AUTISM: INNOVATIONS AND FUTURE DIRECTIONS IN PSYCHOLOGICAL RESEARCH

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EDITORIAL
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Editorial: Autism: Innovations and Future Directions in Psychological Research

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Keywords: autism, psychological research, cognition, perception, neuroscience, participatory research

Editorial on the Research Topic

Autism: Innovations and Future Directions in Psychological Research

Psychological research on autism has a long tradition, covering multiple fields including cognition, perception, clinical research, neuroscience, and social psychology. This Research Topic brings together the latest research in this area, mapping key developments, innovations, and future directions. In this editorial, we will discuss six themes that we have identified across the 22 contributions to this Research Topic: (1) Theories and mechanisms; (2) Characterization of autism; (3) Sensory experiences, perception and movement; (4) Language; (5) Support and interventions; and (6) Methods and technologies. We also provide thoughts on future directions in the field.

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Theories and Mechanisms

Recent discussions have focused on the double-empathy theory (e.g., Milton, 2012; Bolis et al., 2017; but see Georgescu et al., 2020), which interprets communication "difficulties" associated with autism as a bidirectional breakdown between two interaction partners. Building on this theory, Crompton et al. conducted an innovative empirical study examining interpersonal rapport as a function of the neurology of interaction partners, and the person rating levels of rapport. When rating rapport after semi-structured conversations, homogeneous dyads of non-autistic people reported highest levels of rapport, followed by homogeneous dyads of autistic people and lastly mixed (autistic/non-autistic) dyads. Interestingly, taking an outside perspective, when rating observed rapport between interaction partners, homogeneous dyads of autistic individuals were rated highest concerning observed rapport, followed by homogeneous dyads of non-autistic individuals and lastly, again, mixed (autistic/non-autistic) dyads, supporting the double empathy theory.

Beyond specific aspects of functioning, Gernert et al. suggest that empirical and theoretical considerations should move toward a more comprehensive outlook on autism. The authors' Generalized Adaptation Account suggests potential connections between findings from genetics, neurobiology, endocrinology, cellular and neuronal connectivity levels. In this framework, aberrations of neurodevelopmental signaling pathways link up to alterations of neuronal connectivity with cascading effects on neuroendocrine dysregulations and impact on circadian functioning. Consequently, chronic distress and hyperactivation of the hypothalamus-pituitary-adrenal (HPA)-axis result in oxytocinergic downregulation linked to social functioning. This unifying account tries to capture both the complexity of presentation of autism and, in particular, its heterogeneity.

Characterization of Autism

Two articles in this Research Topic were concerned with better characterizing different aspects of autism. Li et al. used the Griffiths Mental Development Scales to characterize the cognitive, motor and social profiles of 398 autistic children (18–96 months old) in China. Findings suggested that many children showed an unbalanced profile (e.g., boys scored better than girls on eye-hand coordination, performance and practical reasoning; and differences in motor behavior became more pronounced with age). Significant aspects to take from this study were the characterization of autistic children in different regions of the world and the need to identify a child's strengths and challenges to develop personalized support.

Characterization can also be useful for predicting the future outcomes of autistic children. Forbes et al. predicted adult outcomes using an impressive dataset of participants who had been repeatedly assessed through childhood, adolescence and adulthood. Only verbal and non-verbal IQ, as well as daily living skills, could be confidently predicted from childhood data while prediction of other aspects (e.g., behavior, adult well-being, depression) was more difficult. Importantly, the authors discuss that views on what constitutes good adult outcomes for autistic children can vary. As acknowledged by the authors, this is clearly a challenging and evolving subject where stakeholder involvement is required.

Sensory Experiences, Perception and Movement

Awareness of the significance of sensory experiences and perceptual processing on the lives of autistic individuals has increased in recent years (Torres and Donnellan, 2015; Autistica, 2016). In this Research Topic, we featured three perceptual studies that all employed rigorous, well-controlled methods to examine this topic. Mihaylova et al. used detailed psychophysical methods to progress understanding of mid-level visual processing in autistic children and adolescents. Results suggested that atypical global grouping (studied in a contour integration task), may be due to higher stimulus-dependent noise in the autistic group, leading to difficulties rejecting background noise and detecting the target.

The effect of low-mid level perceptual differences on higher level perceptual processes was elegantly shown across two studies by Lebreton et al. Here, the authors demonstrated how the commonly reported autistic preference for local compared to global detail impacted upon implicit (unconscious) and explicit (conscious) memory. This is a fascinating finding requiring replication, but has implications for understanding how perceptual style in both autistic and non-autistic individuals affects later memory recall.

Finally, Silver et al. examined whether the intense interests frequently observed in autistic individuals were related to visual processing changes for objects within that category. Contrary to expectations, there were no differences between autistic and non-autistic individuals in visual search abilities for images associated with intense interests. As such, despite enhanced time spent by autistic individuals gazing at images related to an interest, this did not seem to translate to a direct impact on visual processing ability. Linking back to Lebreton et al., we wonder whether the degree of local-global bias in the participants may mediate any relationship between visual experience and visual search ability.

In another fascinating study featured in our Research Topic, Parmar et al. conducted qualitative work with a multidisciplinary team of Optometrists, autism researchers and autistic individuals, using focus groups to provide an in-depth understanding of visual sensory issues. As well as providing a rich description of sensory experiences, the researchers highlighted how visual issues had significant negative impacts on personal well-being and daily life, but also some positive aspects (e.g., detecting details that non-autistic individuals may overlook).

Another article in our Research Topic, by Buckle et al., is the first to highlight Autistic Inertia—a debilitating difficulty of acting on intentions. The article was led by an autistic researcher (based on calls for research on this topic from autistic individuals) and the research highlighted how significant, and potentially common, Autistic Inertia is. Using qualitative methods, the study provided a detailed description of Inertia and the impact of it on autistic people's lives. Two particularly revealing findings were the benefit of other people in helping the individual to overcome being "stuck" and participants wanting to interact with others, but being unable to initiate interaction (which may be interpreted as a lack of social interest).

Language

New approaches in the study of linguistic properties of autism were reported in this Research Topic. Marini et al. combined macrolinguistic (pragmatic, contextual processing) and microlinguistic (word and sentence processing) perspectives of language, which have traditionally been considered independently, showing that morphological and grammatical difficulties were related. Such findings suggest a relationship between difficulties in message planning and organization, which might impact children's grammatical production skills.

New avenues in language research were also highlighted by Sturrock et al. when considering potential gender differences in linguistic studies of autistic people. From a synthesis of previous literature, the authors concluded that there was a very specific profile of language and communication strengths and weaknesses for autistic females without intellectual disability, when compared to both autistic males and non-autistic females. The authors discuss how poorer recognition of autism in females might be influenced by female advantages in aspects of linguistic functioning (but see Lehnhardt et al., 2016).

In a further paper, Williams et al. demonstrated a new approach to studying communication differences between autistic and non-autistic people using relevance theory. This account posits that optimal communication is based on shared and mutually recognized relevance of utterances, which might be mismatched between autistic and non-autistic people when communicating due to differences in experiences of the world. This theoretical approach feeds into the discussions of double-empathy theory (see Theories and mechanisms).

Support and Interventions

Leadbitter et al.'s article proposes that early intervention research could and should be aligned with principles derived from autistic self-advocacy and the neurodiversity movement. Engagement with these principles would lead to, for example, intervention research focusing on changing environments (as opposed to changing autistic people), as well as intervention researchers respecting autistic developmental trajectories and priorities for intervention targets.

In line with this approach, Di Renzo et al. examined the interactions between autistic children and their parents during play, finding that parents who were more accepting of their children's autism diagnosis and who were better able to see things from their children's perspective, were more likely to be attuned with their children during play. Such work highlights the central role of parents as partners in supporting autistic children, and the importance of shared understanding between autistic people and their non-autistic communicative partners (see section Theories and Mechanisms).

Two further studies focused on the important role of parents. Papadopoulos et al. considered support and intervention for young disabled people, 41% of whom had a primary diagnosis of autism. The authors concluded that, to ensure that organized physical activities met the needs of young disabled people, there was a need for activities to be enjoyable, for the participation of siblings and parents to be promoted, and for low-income families to be supported to participate. This work again emphasizes that autism interventions can focus on changing the structures around young people, as opposed to changing the young people themselves.

Relatedly, Devenish et al. examined the effects of lower rates of community participation by autistic young people on their caregivers. Devenish et al. found that if caregivers perceived community supportiveness to be low, this predicted caregiver feelings of isolation. Findings were interpreted within a social model of disability, highlighting how autistic people are disabled by barriers in society.

Not all intervention studies featured in this Research Topic found positive effects of interventions (moving away from the publication bias that once dominated published intervention research). Brehm et al. conducted an initial evaluation of a training programme for parents of autistic children without intellectual/language impairments. The purpose of the evaluation was to evaluate how acceptable the training was for parents, and the results were positive with hardly any parents dropping out of the training programme. Yet a variety of primary outcome measures (e.g., quality of life, social communication) did not show significant improvement. Brehm et al. note that these findings can be useful for directing future work on such interventions.

Similarly, Saul and Norbury presented an alternative to Randomized Controlled Trials for research with rare/complex populations. Drawing on a research study with minimally verbal autistic children, the authors tested the efficacy of a parent-mediated app designed to support speech production, *via* Randomization Tests and Between Case Effect Sizes. As with Brehm et al.'s study, there was no significant effect of the intervention. Yet the research still made an important contribution to the literature; notably demonstrating the importance of robust experimental design and replicable approaches, as well as showing how it is possible to conduct rigorous intervention research with rare or complex samples.

It was also encouraging to see an example of a high-quality case study featured in the article by Courchesne et al., which critically considered the role of interests and strengths in autism, particularly highlighting that these aspects do not necessarily link with academic potential. Courchesne et al. discussed an autistic teenager, C.A., who had above-average musical and calendar calculation abilities, along with pronounced difficulties in other areas (e.g., receptive and expressive language disorder). This discrepancy was found to lead to anxiety, frustration and some behavioral issues due to pressure to use his relative strengths to learn academic skills. Yet, an intervention package that focused on expectations, anxiety and emotional regulation through psychiatric intervention, parental coaching and psychotherapy, improved well-being and behavior. Courchesne et al. caution that while strengths and interests can lead to emotional well-being they should be seen as independent from adaptive outcomes such as academic achievement.

Methods and Technologies

A key message from studies in this theme is the need to develop and validate more ecologically valid assessments of autistic characteristics. For example, Morrison et al. administered standardized measures of social cognition, social skill, and social motivation to autistic and non-autistic adults, and assessed whether these predicted "real-world" social interaction outcomes (measured using unstructured conversations with unfamiliar social partners). While autistic adults scored lower than their non-autistic peers on the three standardized social tasks and were evaluated less favorably during the unstructured social interaction, the links between performance on the standardized measures and unstructured interaction were minimal. The authors therefore question the utility of traditional measures of social performance in autistic people, calling for more ecologically valid assessments.

In line with this approach, Schaller et al. used mobile eye-tracking glasses during autism diagnostic assessments to record gaze behavior of autistic and non-autistic children and adolescents. The authors focused on the percentage of time spent looking at different areas of interest of the face and body of the interviewer and the surrounding space. Significant group differences were found, with non-autistic participants appearing to process faces and facial expressions in a holistic way focusing on the central-face region, whereas autistic participants tended to avoid this face region. The authors stress that the results are preliminary and in need of replication, but this represents an exciting avenue for further work using an ecologically valid methodology.

CONCLUSIONS AND FUTURE DIRECTIONS

Illuminating psychological science on autism from different thematic perspectives has shown several directions we can observe in the field of psychological research. For example: researchers taking a broader perspective, by incorporating previously distinct areas or methods into comprehensive studies; pairing quantitative analysis with qualitative appraisal of experience; putting forward unifying theories spanning different fields; examining an autistic person's strengths and challenges and tailoring more personalized support; developing alternative methods for evaluating interventions in more complex populations; and the implementation of a participatory approach to research. We would like to thank the contributors for their varied and stimulating contributions and hope that this Research Topic stimulates further cutting-edge psychological research that benefits the autistic community.

AUTHOR CONTRIBUTIONS

EG drafted a first version of the Editorial. EG, LC, and CF-W wrote sections of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

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ORIGINAL RESEARCH
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Toward a Definition of the Linguistic Profile of Children With Autism Spectrum Disorder

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The current investigation assessed linguistic and narrative abilities in a cohort of children with Autism Spectrum Disorder (ASD). The linguistic assessment was performed with both traditional tests and a multilevel procedure for discourse analysis. The results showed difficulties at different stages of message planning, organization, and microlinguistic processing (i.e., lexical selection and grammatical processing). Their macrolinguistic impairments were likely related to more general difficulties in the prelinguistic conceptual phase of message planning and mental model generation. Such weaknesses included a difficulty in the non-verbal conceptualization of the story and the generation of an internal representation of the addressee's mental model.

Keywords: Autism Spectrum Disorder, language, narrative analysis, developmental neuropsychology, neurolinguistics

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INTRODUCTION

Autism spectrum disorder (ASD) is characterized by persistent deficits in social communication and interaction associated with restricted and repetitive patterns of behavior, interests or activities (American Psychiatric Association, 2013). Because of its pivotal role in communicative interactions, since the seminal descriptions provided by Kanner (1943) and Asperger (1944/1991) language development and functioning in ASD has been the focus of extensive research (see also Boucher, 2012 for a comprehensive review). However, an accurate linguistic assessment of individuals with ASD must consider the actual complexity of the linguistic system. Language can be assessed from a micro- and a macrolinguistic perspective (Glosser and Deser, 1990; Marini et al., 2011): the microlinguistic perspective focuses on the intra-sentential (i.e., within-utterance) organization of discourse by assessing the phonetic, phonological and morphological skills needed to process words (lexical processing) and the morphosyntactic and syntactic abilities involved in the generation of sentences (syntactic processing); the macrolinguistic perspective focuses on the inter-sentential (i.e., between-utterances) processing by assessing the ability to select contextually appropriate words and utterances (pragmatic processing) and to generate cohesive and coherent ties among the sentences (discourse processing; Kintsch, 1994).

Longitudinal studies on language development in ASD have shown that the linguistic profiles of these children might change significantly with age (Bennett et al., 2008; Geurts and Embrechts, 2008; Rapin et al., 2009). Preschoolers are most likely to show phonological (but not necessarily articulatory) and grammatical impairments. For example, Tuchman et al. (1991) reported that in a cohort of 197 children with ASD, 117 individuals (59%) showed phonological and grammatical difficulties. Similarly, Allen and Rapin (1992) showed that all the individuals in a cohort of

229 preschoolers with ASD aged between 4 and 5 years showed not only pragmatic impairments but also some difficulties in linguistic comprehension. Sixty-three percent of these children (N = 144) experienced also phonological and syntactic impairments. These two studies had partially overlapping cohorts of individuals. However, grammatical impairments have been observed also in different groups of preschoolers with ASD (e.g., Eigsti et al., 2007). If phonological and grammatical impairments are frequent in preschoolers, pragmatic disturbances predominate by school-age (Geurts and Embrechts, 2008).

While informative and interesting, studies on language in ASD have usually focused on single aspects of language processing without considering it in its complexity. At times, this has led to mixed results. Indeed, even if pragmatic and discourse difficulties are a common finding in individuals with ASD, not all of them show phonological, lexical and/or grammatical difficulties. Even those who experience these symptoms may exhibit large within-group variability (e.g., Rapin and Dunn, 2003). In the 80's, such observations led to the exclusion of language impairments from the criteria for the diagnosis of ASD and prompted a gradual shift of attention from the description of the linguistic features of the general population of individuals with ASD taken as a whole to more focused analyses of the linguistic characteristics of specific subgroups with linguistic impairment or delay (e.g., Kjelgaard and Tager-Flusberg, 2001; Tager-Flusberg and Joseph, 2003; Whitehouse et al., 2008). For example, Kjelgaard and Tager-Flusberg (2001) administered a range of language tests to 89 children with ASD aged 4–14 years. They were highly heterogeneous. Indeed, according to their performance on a test of lexical comprehension, the authors managed to cluster them in three major subgroups: one with normal linguistic performance (Autistic individuals with Normal Language, ALN); one with borderline performance and scores ranging within 1 and 2 standard deviations below the mean (Autistic individuals with borderline language skills); one with overtly impaired performance (Autistic individuals with Language Impairment, ALI). Articulation was normal in the ALN and borderline subgroups and mildly impaired in the ALI population. However, all three subgroups experienced difficulties on tasks assessing lexical comprehension and production (especially the ALI population) with the most important difficulties on tasks assessing pragmatic skills. Importantly, in this study the performance on a task of non-word repetition proved highly sensitive to the presence of linguistic disturbances: only the ALI subgroup was found significantly impaired. Subsequent studies focusing on school-age children with ALI ranging from 6 years up highlighted persisting morphological difficulties (Roberts et al., 2004) often characterized by the omission or substitution of function words (e.g., prepositions, articles or conjunctions; Lai, 2011). In spontaneous language, these difficulties may lead to reduced mean length of utterance (MLU; measured in morphemes as in Condouris et al., 2003) and syntactic structures that are fewer (e.g., Lai, 2011) and less variable (e.g., Losh and Capps, 2003) than normal. This is interesting, as often no syntactic difficulties are noticeable when their grammatical skills are assessed in more structured and decontextualized tests (e.g., Shulman and Guberman, 2007).

Traditional tests cannot adequately describe the linguistic profile of children with communicative disorders (e.g., Marini et al., 2008; Volden et al., 2017). To capture the interactions between different linguistic skills, it is necessary to include also procedures of narrative discourse assessments (Marini et al., 2005, 2014). Indeed, the generation of an informative message requires the speaker to consider the context and tie the different propositions through cohesive and coherent links. Therefore, a comprehensive assessment cannot be limited to the analysis of the microlinguistic features of message production but must include also the macrolinguistic ones (Volden et al., 2017). Overall, the narrative language produced by children with ASD has been described as idiosyncratic at both micro- and macrolinguistic levels of processing (e.g., Baixauli et al., 2016). Microlinguistic difficulties include the production of utterances characterized by unusual words, aberrant prosodic contours, and instances of pronoun reversal (e.g., Kuijper et al., 2017) with anomalous productivity levels and grammatical structuring (Baixauli et al., 2016). Macrolinguistic difficulties include the production of speech samples that are perceived as contextually inappropriate for the inclusion of echolalic, repetitive and overtly incoherent utterances (e.g., Kuijper et al., 2017). Furthermore, they have significant difficulties in the production of appropriately informative referring expressions (e.g., Arnold et al., 2009; Banney et al., 2015). As to this regard, a recent investigation by Malkin et al. (2018) showed that, even if they can take to some extent the interlocutor-specific prior experience into account, children with ASD may lag behind typical peers in the degree to which they make use of such information. Difficulties have been reported in the ability to establish causal connections between the utterances (e.g., Diehl et al., 2006; Baixauli et al., 2016; Volden et al., 2017) and organize the temporal dynamics of narrative discourse (Ferretti et al., 2018; Marini et al., 2019) to the extent that they are often not able to adequately use story-grammar information to organize their narrative speech samples (Goldman, 2008; McCabe et al., 2013).

As it is evident from this brief analysis of the available literature, linguistic skills in ASD have been widely explored. However, some issues remain unresolved. First, it is not clear yet whether a morphological difficulty can be ascribed to persons with ASD and language impairment and whether it is related to their grammatical skills while producing a narrative discourse. For example, the already mentioned study by Roberts et al. (2004) suggests that difficulties in verb tense might be an important marker of the linguistic symptomatology observable in these children. Evidence of morphological difficulties leading to omissions of function words further supports this possibility (e.g., Botting and Conti-Ramsden, 2003; Condouris et al., 2003). However, to the best of our knowledge, no study has explicitly explored the possible relation between the morphological impairments often observed in ALI children and grammatical (i.e., morphosyntactic and syntactic) difficulties in discourse production. In our view, one further aspect requires explicit analysis: the possibility that different types of macrolinguistic difficulties are related to the microlinguistic impairments observable on a narrative production task. Consequently, this study aimed to replicate and expand upon previous research on both micro- and

macrolinguistic skills in a group of Italian-speaking school-age children with ASD and microlinguistic impairment (ALI). Namely, to have a detailed profile of their linguistic and narrative skills we jointly adopted traditional standardized procedures for linguistic analysis and a multilevel procedure for discourse analysis that has proven useful in detecting micro- and macrolinguistic impairments in both children and adult patients with communicative disorders (e.g., Marini et al., 2010, 2014). We assumed that this accurate analysis would allow us to efficiently describe the micro- and macrolinguistic abilities of the children with ALI and provide additional information about these features in children with a language, Italian, that is structurally dissimilar from English. Furthermore, as it enables the exploration of the complex interactions between micro- and macrolinguistic processes, we hypothesized that the multilevel procedure for discourse analysis would highlight the potential effect of macrolinguistic variables on microlinguistic (i.e., lexical and sentence-level processing) performance. In particular, it was hypothesized that selective problems in microlinguistic processing would be related to a more general problem in discourse planning and organization.

METHODS

Participants

Seventy-four Italian-speaking participants were included in the study. They formed an experimental and control group. The experimental cohort consisted of 24 children with ASD aged between 7 and 11;11 years old (mean 9 years and 3 months; standard deviation, SD, 1.70). They had been diagnosed by expert clinicians. Inclusion criteria included the absence of intellectual disability, brain lesions, or auditory difficulties (see **Table 1**) but the presence of language impairments as certified by a speech therapist and a performance of at least 1.5 standard deviations below expected means on a test of Non-Word Repetition (Marini et al., 2015). Therefore, all participants with ASD had linguistic impairment (ALI).

The control cohort included 50 participants with Typical Language Development (TLD) aged between 7 and 11;11 years old (mean 9 years and 0 months; SD 1.51). They were selected in order to roughly match two controls for every participant with ASD. Inclusion criteria included a normal performance on Raven's progressive matrices (Raven, 1938), the non-word repetition subtest of the PROMEA (Vicari, 2007), and on the forward and backward digit spans subtests of the Wechsler Scales (Wechsler, 1993). No learning or language difficulties were reported.

The two groups did not differ on age, education or on performance at Raven's progressive matrices (see **Table 1**). As expected, an independent-samples *t*-test confirmed that the cohort with ASD scored lower than controls on the Non-Word Repetition subtest of the "Batteria per la Valutazione del Linguaggio in Bambini dai 4 ai 12 anni" (BVL_4-12, Marini et al., 2015) [$t(46) = -5.873$; $p < 0.001$]. All participants came from middle-class families. The study received institutional ethics approval by the ethics committee of the Research Institute IRCCS "E. Medea". All parents released their informed consent to the participation of their children to the study and the treatment of the data.

TABLE 1 | Means (and standard deviations) showing demographic data of the two groups of participants and their performance on the Raven's colored matrices and on the Non-word Repetition task.

	ASD (*N* = 24)	TLD (*N* = 50)
Age	9.25 (1.70)	8.65 (1.54)
Education	3.83 (1.90) – range: 1st–6th grade	3.42 (1.53) – range: 1st–6th grade
Raven	23.25 (8.28)	27.82 (4.27)
Non-word repetition*	12.04 (2.12)	14.70 (0.54)

The asterisk shows when the group-related difference was significant ($p < 0.05$). ASD, children with Autism Spectrum Disorder; TLD, children with Typical Language Development.

Procedures of Linguistic Assessment

The linguistic assessment was delivered by trained speech-therapists or developmental psychologists in a quiet room at the Research Centers "Ospedale Pediatrico Bambin Gesù" and "E. Medea" (for children with ASD) or their schools (for children with TD). The linguistic assessment focused on lexical, grammatical, and macrolinguistic skills.

Assessment of Lexical Skills

The children's lexical skills were assessed by administering tasks focusing on lexical production and comprehension. Namely, the children received three subtests of the BVL_4-12 assessing naming, lexical comprehension, and discourse production.

In the **naming** task, children are required to name 67 drawings referring to 51 nouns (divided into 15 semantic categories) and 16 action verbs. These words were carefully selected for their frequency of use in Italian (Very high: 17; High: 23; Low: 27). Each correct answer is assigned 1 point. The maximum score is 67.

In the **lexical comprehension** task participants are required to identify which, among four pictures, best represents the meaning of the word produced by the examiner. The pictures represent a target word (i.e., the meaning of the word produced by the examiner, for example, "cat"), a semantic distracter (e.g., a picture portraying the meaning of a word which is semantically related to the target word; in this case "dog"), a phonological distracter (e.g., a picture portraying the meaning of a word which is phonologically related to the target word; "car"), and an unrelated distracter (e.g., "table"). All target words (31 nouns, 10 verbs, and 1 adjective) have been selected according to their frequency in Italian (4 items with very high, 8 with high, and 30 with a low frequency of use). Each correct answer is assigned 1 point for a maximum score of 42.

The **narrative assessment** was performed by analyzing the speech samples obtained by administering the "Nest Story" description task (Paradis, 1987). The recordings of the story descriptions were transcribed and analyzed by two independent coders according to the procedures described

in Marini et al. (2011). Namely, the analysis focused on the participants' speech rates and percentages of semantic errors, paragrammatic errors, omissions of function words, complete sentences, local and global coherence errors, and lexical and thematic informativeness (please see **Appendix A** for an example of the scoring procedure). The scoring procedure was performed independently by two raters and then compared. The raters were blind with respect to the fact that the transcripts related to stories produced by children with ASD or TD. An inter-rater reliability analysis using the Kappa statistic was performed to determine consistency among raters. Acceptable inter-rater reliability was defined as $k \geq 0.80$ (Carletta, 1996; Marini and Urgesi, 2012). The interrater reliability scores for the two raters were constantly high. During the analysis, in a few cases the scorers needed to listen again to the audio recordings to face the residual minor issues that could be easily solved.

As for the assessment of their lexical skills, the analyses focused on Speech Rate (words per minute) and the percentage of Semantic Errors. The **Speech rate** was calculated by dividing the number of words produced by the child by the time spent during narrative production (in seconds, using the following formula: (Words/Time_in_seconds)*60. Semantic errors were assessed in terms of both semantic and verbal paraphasias. A semantic paraphasia was scored whenever a target word had been replaced by a semantically related one [e.g., *Fiore* (in English: *Flower*) instead of *Albero* (in English: *Tree*)]. A verbal paraphasia was scored if the target word had been replaced by a semantically unrelated one [e.g., *Cane* (in English: *Dog*) instead of *Albero* (in English: *Tree*)]. The **percentage of Semantic Errors** was calculated by summing semantic and verbal paraphasias and dividing this value by the number of words produced during the narrative description. This score was multiplied by 100.

Assessment of Morphological and Grammatical Skills

The assessment of morphological and grammatical skills included tasks focusing on morphological and grammatical production and comprehension skills. Namely, the children received three subtests of the BVL_4-12 assessing sentence completion, syntactic comprehension, and narrative production.

In the **sentence completion** task children are required to produce grammatically sound sentences by processing verbal derivational and inflectional morphology. After hearing a sentence that provides a model [e.g., *Marco apre la porta* (in English: *Marco opens the door*)], the child is presented with the beginning of a second one [the prompt; e.g., *Anche noi* . . . (in English: *We also*. . .)] that (s)he is asked to complete assigning the correct morphemes to the verb [the target; e.g., *Anche noi apriamo la porta* (in English: *We also open the door*)]. The test is made of 14 pairs of model sentences and prompts with different levels of grammatical complexity. The first five sentences assess the ability to process inflective morphology with bound morphemes (e.g., *apriamo*). From the sixth item children are asked to cope with more complex sentences with the use of both derivational and inflective morphology [e.g., Model – *Oggi Maria è aiutata dalla mamma a fare i compiti* (in English: *Today, Maria is helped by her mother to do her homework*); Prompt: *Anche ieri Maria* . . . (in English: *Even yesterday, Maria* . . .); Expected response: *Anche ieri Maria è stata aiutata dalla mamma a fare i compiti* (in English: *Even yesterday, Maria was helped by her mother to do her homework*)]. Each correct answer is assigned 1 point with a maximum score of 14.

In the test of **syntactic comprehension**, participants are asked to match each of 40 sentences of increasing grammatical complexity with one of four pictures. The pictures represent the meaning of the sentence uttered by the examiner (the target) and three distracters referring to alternative sentences that differ from the target for the presence of inverted thematic roles or other morphosyntactic alterations. For example, after hearing the sentence *Il bambino che è in bicicletta rincorre la bambina che è a piedi* (in English: *The boy who's on a bike chases the girl who's on foot*), the child is shown a sheet with four pictures: one depicting its meaning (target) and three distracters representing: 1. *The girl who's on a bike chases the boy who's on foot*; 2. *The boy who's on a bike chases the girl who's on a bike*; 3. *The girl who's on a bike is beside the boy who's on foot*. Each correct answer is assigned 1 point with a maximum score is 40.

The narrative assessment allowed us to obtain a % of Paragrammatic Errors to words, a % of Omission of Function Words to utterances and a % of Complete Sentences. Paragrammatic errors reflect morphological difficulties that include a misuse of bound morphemes {e.g., **Questo è una signora* [in English: "**this* (masculine) *is a woman* (feminine)"]} and/or function words [e.g., *Il signore sale *con l'albero* (in English: "*The man climbs *with the tree*")]. The **%Paragrammatic Errors** was calculated by dividing the number of Paragrammatic errors by the number of words and multiplying this value by 100. An omission of function words was scored whenever a child omitted fa unction word that was necessarily requested by the sentence [e.g., *Ramo si spezza (in English: "*Branch breaks"]. The **% Omission of Function Words** was calculated by dividing the number of such omissions by the number of utterances and multiplying this value by 100. As for the **% of Complete Sentences**, a sentence was considered grammatically complete if all of the arguments required by the verb had been inserted correctly and if no omissions or substitutions of free or bound morphemes were detectable. Therefore this percentage was calculated by dividing the number of complete sentences by the number of utterances and then multiplying this value by 100.

Assessment of Macrolinguistic Skills

The macrolinguistic skills were assessed in terms of textual organization and informative content. The former aspect was accounted for by calculating a % of Local Coherence Errors and a % of Global Coherence Errors. Local coherence errors were calculated in terms of topic shifts (occurring when an utterance was abruptly interrupted and the following one introduced new information instead of completing the one left incomplete; e.g., / the man is staring at . . . / and here he is falling /, where the first utterance remained incomplete as the second one introduced a new argument) and missing referents (i.e., instances of words whose referent was not clear or missing as in the following example: /Here they look at a nest / He climbs. . ./). The **% of Local Coherence Errors** was calculated by summing instances of

topic shifts and missing referents, dividing them by the number of utterances in the speech sample and multiplying this value by 100.

Global coherence errors were calculated in terms of utterances that were tangential, conceptually incongruent with the story, repetitions or simple fillers (see Marini et al., 2011 for a detailed description of such errors). The **% of Global Coherence Errors** was calculated by summing instances of tangential, incongruent, repetitive and filler utterances, dividing them by the number of utterances in the speech sample and multiplying this value by 100.

The informative content of the narrative descriptions was assessed in terms of lexical and thematic informativeness. Lexical informativeness was calculated by counting the amount of Lexical Information Units, i.e., those words that were appropriate from a phonological, grammatical and pragmatic point of view. Hence, phonological, morphological and semantic errors, as well as words contained in tangential, repetitive, filler, or semantically incongruent utterances, were excluded from this count. The **% of Lexical Informativeness** was calculated by dividing the number of lexical information units by the number of words produced during the storytelling and multiplying this value by 100.

Finally, the **% of Thematic Informativeness** for each story was measured by dividing the number of thematic units (i.e., those elements of content portrayed in the picture stimulus) produced in each story by the total amount of thematic units available in that story and multiplying this value by 100.

RESULTS

Assessment of Lexical Skills

The Levene's test for equality of variances showed that the assumption of homogeneity of variance had been violated for measures of Naming ($p < 0.001$), Lexical Comprehension ($p < 0.001$), Speech Rate ($p < 0.010$), and % Semantic Errors ($p < 0.001$). For this reason, non-parametric Mann–Whitney tests were used to explore between-subject effects on these measures (see **Table 2**). The level of statistical significance was set at $p < 0.013$ (0.05/4 dependent variables) after Bonferroni correction for multiple comparisons. The group with ASD showed difficulties in all these variables: Naming ($U = 292.00$; $p < 0.001$); Lexical Comprehension ($U = 371.50$; $p < 0.008$); Speech Rate ($U = 313.50$; $p < 0.001$); and % Semantic Errors ($U = 297.00$; $p < 0.001$). Considering a z-score of -1.5 as a cut-off for normality for Speech Rate, Lexical Comprehension and Naming and +1.5 for the production of Semantic Errors, a significant number of participants with ASD scored below normal range in these lexical variables (see **Figure 1**): 46% in Naming (8% scored -1.5; 38% scored -2); 42% in Lexical Comprehension (4% scored -1.5; 38% scored -2); 25% in Speech Rate (4% scored -1.5; 21% scored -2); 59% in % Semantic Errors (13% scored -1.5; 46% scored -2).

TABLE 2 | Results of the analysis of lexical skills in the groups of participants with ASD and TLD.

Assessment of lexical skills	ASD	TLD
Naming*	54.21 (8.76)	61.24 (3.81)
Lexical comprehension*	30.71 (8.11)	35.96 (3.46)
Speech rate*	84.10 (41.91)	103.06 (22.24)
% Semantic errors*	2.45 (3.08)	0.39 (0.67)

Asterisks show when the group-related difference was significant after Bonferroni correction for multiple comparisons ($p < 0.013$). ASD, children with Autism Spectrum Disorder; TLD, children with Typical Language Development.

Assessment of Grammatical Skills

The Levene's test for equality of variances showed that the assumption of homogeneity of variance had been violated for the measures assessing grammatical skills: Sentence Completion ($p < 0.001$), Syntactic Comprehension ($p < 0.001$), % Paragrammatic Errors ($p < 0.001$), % Omissions of Function Words ($p < 0.001$) and % Complete Sentences ($p < 0.001$). For this reason, a series of non-parametric Mann–Whitney tests with Group (ASD vs. TLD as fixed factor) and the grammatical measures as dependent variables were used to explore between-subject effects (see **Table 3**). The level of statistical significance was set at $p < 0.010$ (0.05/5 dependent variables) after Bonferroni correction for multiple comparisons. These analyses showed that participants with ASD performed worse than healthy peers in Syntactic Comprehension ($U = 295.00$; $p < 0.001$), Sentence Completion ($U = 214.00$; $p < 0.001$), % Paragrammatic errors ($U = 293.00$; $p < 0.001$), % Omissions of Function Words ($U = 339.50$; $p < 0.001$), and % Complete Sentences ($U = 312.50$; $p < 0.001$). Considering a z-score of -1.5 as a cut-off for normality for Sentence Completion, Syntactic Comprehension, % Paragrammatic Errors and % Complete Sentences and +1.5 for the production of Paragrammatic Errors (normative data for the % of Omissions of Function Words were not available), the majority of participants with ASD scored well below normal range in most of these grammatical variables (see **Figure 2**): 71% in Sentence Completion (8% scored -1.5; 63% scored -2); 67% in Syntactic Comprehension (25% scored -1.5; 42% scored -2); 51% in % Complete Sentences (13% scored -1.5; 38% scored -2); 54% in % Paragrammatic Errors (25% scored +1.5; 29% scored +2).

Assessment of Macrolinguistic Skills

As the Levene's test for equality of variances showed that the assumption of homogeneity of variance had been violated for measures assessing % of Errors of Local ($p < 0.001$) and Global Coherence ($p < 0.001$) and % of Lexical Informativeness ($p < 0.001$) but not for % Thematic Selection ($p = 0.833$), group-related differences on such measures were analyzed with three Mann–Whitney tests for % of Errors of Local and Global Coherence and % of Lexical Informativeness and one independent-samples *t*-test for % Thematic Selection (see **Table 4**). The level of statistical significance was set at $p < 0.013$ (0.05/4 dependent variables) after Bonferroni correction for multiple comparisons. **Table 4** reports the results of these analyses. Overall, the group of participants with ASD produced more errors of Local ($U = 211.00$; $p < 0.001$) and Global Coherence ($U = 246.50$; $p < 0.001$), their narrative samples were characterized by lower levels of lexical informativeness ($U = 220.00$; $p < 0.001$) and Thematic Selection [$t(72) = -5.493$; $p < 0.001$)]. Considering a z-score of -1.5 as a

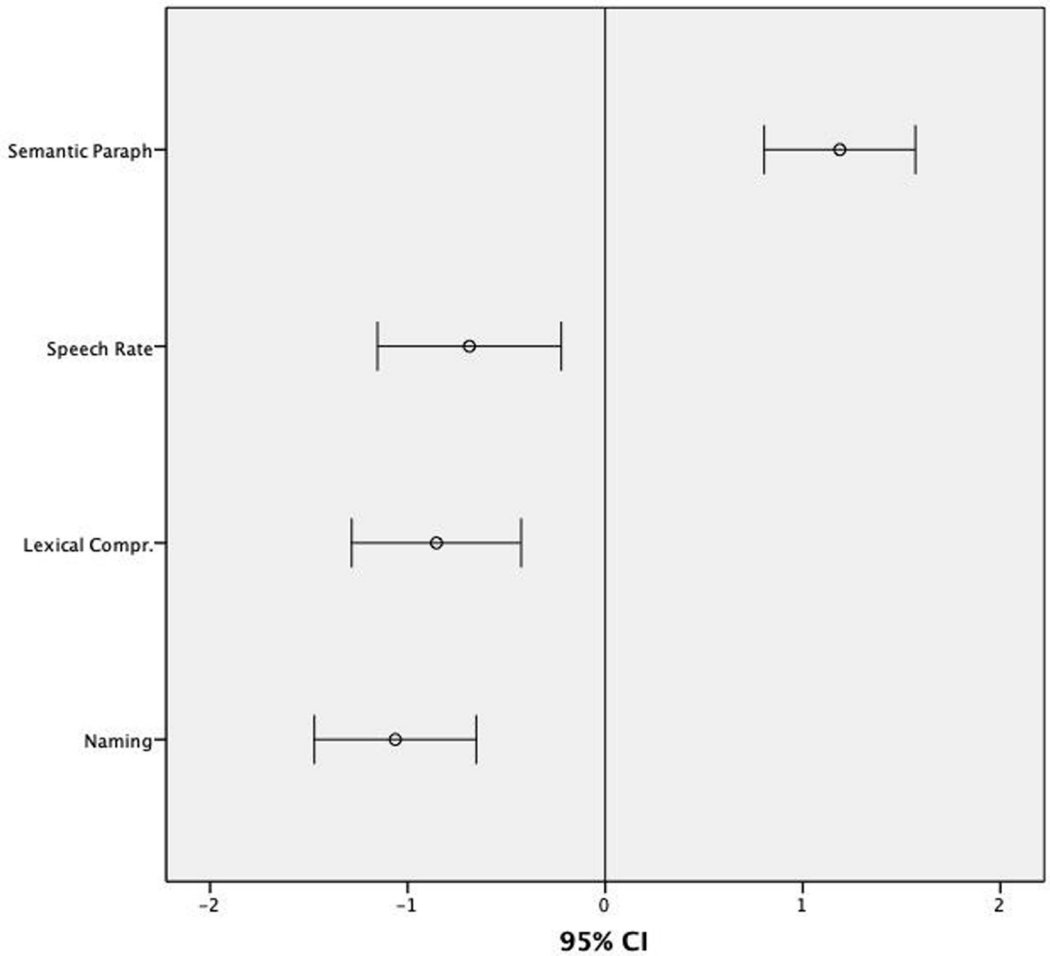

FIGURE 1 | Mean z-scores and 95% CIs for the scores assessing lexical skills in children with ASD in relation to normative data.

cut-off for normality for % Lexical Informativeness and +1.5 for the production of Local Coherence Errors and Global Coherence Errors (normative data for the % Thematic Selection were not available), the majority of participants with ASD scored well below normal range in most of these macrolinguistic variables (see **Figure 3**): 63% in % Lexical Informativeness (21% scored -1.5; 42% scored -2); 67% in % Local Coherence Errors (13% scored +1.5; 54% scored +2); 55% in % Global Coherence Errors (13% scored +1.5; 42% scored +2).

Reassessment of Group Related Differences After Balancing the Two Groups for Number of Participants

As stated in section Participants, the control participants were selected in order to roughly match two controls for every participant with ASD. While providing a quite robust comparison with linguistic skills in children with TLD, this choice might have biased our results because of an unequal number of participants in the two groups. For this reason, the same analyses described in sections Assessment of Lexical Skills, Assessment of Grammatical Skills, and Assessment of Macrolinguistic Skills were re-run after reducing the cohort of control participants by selecting them on the base of their age in order to roughly match one control for every participant with ASD (please see **Table 5** for the mean demographic data of the reduced control sample and their performance on the Raven's colored matrices and on the Non-word Repetition task).

TABLE 3 | Results of the analysis of grammatical skills in the groups of participants with ASD and TLD.

Assessment of grammatical skills	ASD	TLD
Sentence completion*	7.38 (3.92)	11.86 (1.92)
Syntactic comprehension*	30.96 (7.06)	36.18 (2.17)
% Paragrammatic errors*	3.11 (4.04)	0.42 (0.78)
% Omissions of function words*	14.59 (24.58)	0.83 (2.67)
% Complete sentences*	44.34 (29.92)	64.68 (16.70)

Asterisks show when the group-related difference was significant after Bonferroni correction for multiple comparisons ($p < 0.010$). ASD, children with Autism Spectrum Disorder; TLD, children with Typical Language Development.

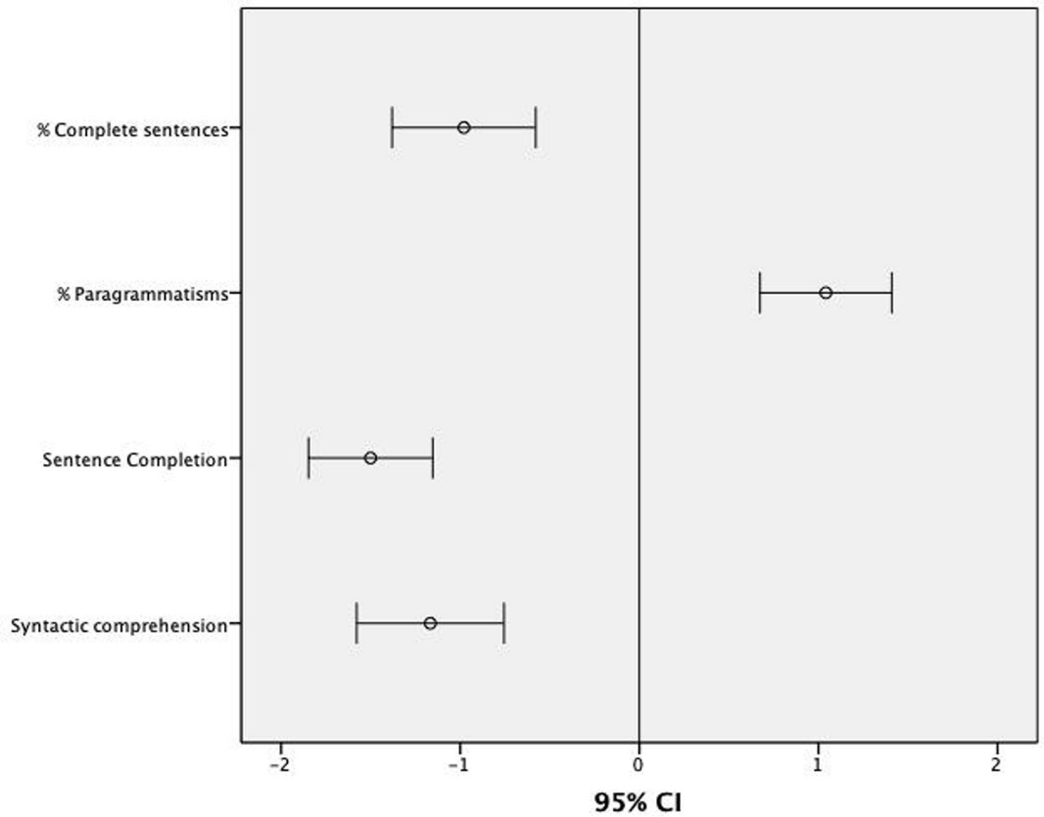

FIGURE 2 | Mean z-scores and 95% CIs for the scores assessing grammatical skills in children with ASD in relation to normative data.

TABLE 4 | Results of the analysis of textual organization and informative content on the narrative production task in the groups of participants with ASD and TLD.

Analysis of textual construction and informative Content	ASD	TLD
% Local coherence errors*	38.81 (31.17)	8.28 (8.61)
% Global coherence errors *	23.49 (15.79)	7.72 (8.77)
% Lexical informativeness*	62.00 (22.19)	83.05 (9.82)
%Thematic selection*	21.18 (12.59)	37.67 (11.84)

Asterisks show when the group-related difference was significant after Bonferroni correction for muplitple comparisons ($p < 0.013$). ASD, children with Autism Spectrum Disorder; TLD, children with Typical Language Development.

Lexical Skills

Levene's test for equality of variances showed that the assumption of homogeneity of variance had been violated for measures of Naming ($p < 0.001$), Lexical Comprehension ($p < 0.001$), Speech Rate ($p < 0.016$), and % Semantic Errors ($p < 0.001$). For this reason, non-parametric Mann–Whitney tests were used to explore between-subject effects on these measures (see **Table 6**). Statistical significance was set at $p < 0.013$ (0.05/4 dependent variables) after Bonferroni correction for multiple comparisons. The group with ASD showed difficulties in all these variables: Naming ($U = 120.50$; $p < 0.001$); Lexical Comprehension ($U = 138.00$; $p < 0.002$); Speech Rate ($U = 111.00$; $p < 0.001$); and % Semantic Errors ($U = 135.00$; $p < 0.001$).

Grammatical Skills

Levene's test for equality of variances showed that the assumption of homogeneity of variance had been violated for the measures assessing grammatical skills: Sentence Completion ($p < 0.001$), Syntactic Comprehension ($p < 0.001$), % Paragrammatic Errors ($p < 0.001$), % Omissions of Function Words ($p < 0.001$), and % Complete Sentences ($p < 0.001$). For this reason, non-parametric Mann–Whitney tests with Group (ASD vs. TLD as independent variable) and the grammatical measures as dependent variables were used to explore between-subject effects (see **Table 6**). Statistical significance was set at $p < 0.010$ (0.05/5 dependent variables) after Bonferroni correction for multiple comparisons. These analyses revealed that individuals with ASD performed worse than healthy peers in Syntactic Comprehension ($U = 112.50$; $p < 0.001$), Sentence Completion ($U = 78.50$; $p < 0.001$), % Paragrammatic errors ($U = 145.00$; $p < 0.002$), % Omissions of Function Words ($U = 166.50$; $p < 0.002$), and % Complete Sentences ($U = 147.00$; $p < 0.004$).

Macrolinguistic Skills

As Levene's test for equality of variances showed that the assumption of homogeneity of variance had been violated

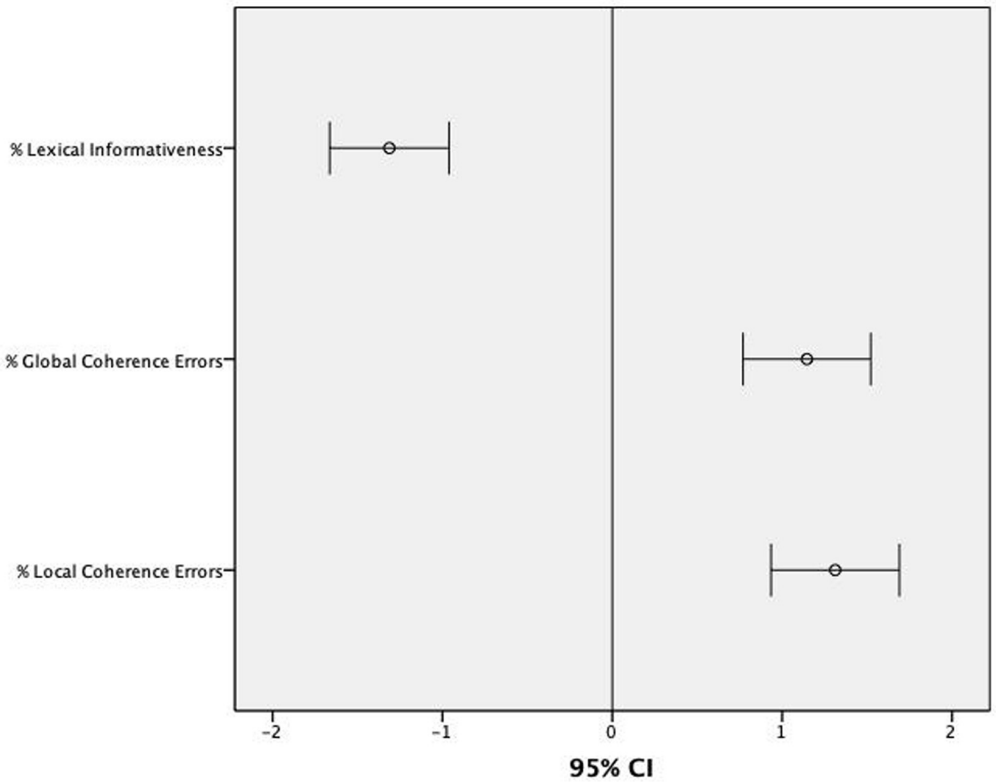

FIGURE 3 | Mean z-scores and 95% CIs for the scores assessing narrative skills in children with ASD in relation to normative data.

TABLE 5 | Means (and standard deviations) showing demographic data of the group of controls after the reduction to equal the number of participants in the two groups.

	TLD (*N* = 24)
Age	9.05 (1.51)
Education	3.83 (1.52) – range: 1st–6th grade
Raven	28.54 (4.44)
Non-word repetition*	14.67 (0.12)

*Their performance on the Raven's colored matrices and on the Non-word Repetition task are also reported. The asterisk shows when the group-related difference was significant ($p < 0.05$). For scores of the participants with ASD please refer to **Table 1**. TLD, children with Typical Language Development.*

TABLE 6 | Results of the analysis of lexical, grammatical, and macrolinguistic skills in the reduced group of participants with Typical Language Development.

Lexical skills	
Naming*	62.17(2.63)
Lexical comprehension*	37.04(2.85)
Speech rate*	112.01(17.63)
% Semantic errors*	0.29(0.58)
Grammatical skills	
Sentence completion*	12.54(1.02)
Syntactic comprehension*	36.96(1.81)
% Paragrammatic errors*	0.44(0.61)
% Omissions of function words*	0.94(2.57)
% Complete sentences *	65.48(12.75)
Macrolinguistic skills	
% Local coherence errors*	6.34(6.84)
% Global coherence errors *	6.99(7.63)
% Lexical informativeness*	85.24(6.78)
% Thematic selection*	42.01(13.68)

*Asterisks show when the group-related difference was significant after Bonferroni correction for multiple comparisons. For scores of the participants with ASD please refer to **Tables 2–4**.*

for measures assessing % of Errors of Local ($p < 0.001$) and Global Coherence ($p < 0.001$) and % of Lexical Informativeness ($p < 0.001$) but not for % Thematic Selection ($p = 0.532$), group-related differences on such measures were analyzed with three Mann–Whitney tests for % of Errors of Local and Global Coherence and % of Lexical Informativeness and one independent-samples *t*-test for % Thematic Selection (see **Table 6**). Statistical significance was set at $p < 0.013$ (0.05/4 dependent variables) after Bonferroni correction for multiple comparisons. As reported in **Table 4**, the group of participants with ASD produced more errors of Local ($U = 87.50$; $p < 0.001$) and Global Coherence ($U = 110.50$; $p < 0.001$), their narrative samples were characterized by lower levels of lexical informativeness ($U = 85.00$; $p < 0.001$) and Thematic Selection [$t(46) = -5.491$; $p < 0.001$].

Do Morphological Difficulties Relate to Grammatical Impairments in Children With ASD and Controls?

A goal of this study was to determine whether the morphological difficulties often observed in children with ASD are related to grammatical (i.e., morphosyntactic and syntactic) difficulties while producing samples of narrative language. This was explored by performing a series of correlational analyses between the children's performance on the test assessing Sentence Completion and the other grammatical variables obtained with traditional tasks (i.e., Syntactic Comprehension) and narrative analysis (i.e., % Omission Function Words and % Complete Sentences). These analyses showed that, in children with ASD, the performance on the sentence completion task correlated with all of the above-mentioned variables: Syntactic Comprehension (Spearman's Rho = 0.797; $p < 0.001$), % Complete Sentences (Spearman's Rho = 0.517; $p < 0.010$), % Omission Function Words (Spearman's Rho = -0.483; $p < 0.017$).

On the contrary, the performance of participants with TLD on the sentence completion task did not correlate with any of the above-mentioned variables: Syntactic Comprehension (Spearman's Rho = 0.002; $p = 0.992$), % Complete Sentences (Spearman's Rho = -0.395; $p = 0.056$), % Omission Function Words (Spearman's Rho = -0.057; $p = 0.793$).

Are Macrolinguistic Disturbances Related to Microlinguistic Difficulties in Children With ASD and Controls?

The possibility that macrolinguistic disturbances (i.e., % of Local and Global Coherence Errors) might be related to the microlinguistic difficulties (i.e., measures of lexical and grammatical skills) was explored by using Spearman's Rho correlation coefficient. In the group of children with ASD both % Global and % Local Coherence Errors were negatively correlated to the % Complete Sentences (Global Coherence Errors: Spearman's Rho = -0.477; $p < 0.018$; Local Coherence Errors: Spearman's Rho = -0.430; $p < 0.036$).

On the contrary, in participants with TLD the % Complete Sentences did not correlate with the production of Global (Spearman's Rho = -0.193; $p = 0.366$) or Local Coherence Errors (Spearman's Rho = -0.070; $p = 0.745$).

DISCUSSION

This study investigated linguistic and narrative abilities in a cohort of children with ASD and language impairments. The linguistic assessment was performed with both traditional tests and a multilevel procedure for discourse analysis. Overall, analyses involving both the complete sample of participants ($N = 74$ with 50 controls) and the reduced number of participants ($N = 48$ with 24 controls) showed that the children with ASD had significant lexical, grammatical and narrative difficulties. A series of correlational analyses confirmed that (1) morphological difficulties were related to the observed grammatical impairments; (2) global coherence errors were negatively correlated to the production of complete sentences.

Not surprisingly, the participants with ASD and language impairments showed significant narrative difficulties. Indeed, 67% and 65% of them produced a significant amount of local and global coherence errors (see also Kuijper et al., 2017) that likely contributed to the reduction of their levels of lexical informativeness. Indeed, their narratives were characterized by the inclusion of repetitive and overtly incoherent utterances that were quite often not correctly linked with each other (e.g., Diehl et al., 2006; Baixauli et al., 2016; Volden et al., 2017). Of note, they also produced fewer ideas that were portrayed in the vignettes as reflected by the reduced % of Thematic Selection. This last finding may suggest that a significant difficulty was in the phase of non-verbal conceptualization of the story. According to the Structure Building Framework (Gernsbacher, 1990; see Marini et al. (2017) for its application in the domain of narrative production) the generation of a narrative discourse relies on a multistage process. In the first stage, prelinguistic, it is necessary to generate a mental model or scenario of the story that will serve as a foundation for its development. As the information flows, the speaker needs to continuously monitor the consistency of such mental models and scenarios with the generated structures. In case of inconsistency, it becomes necessary to generate new structures that are in line with the desired mental model. These will eventually trigger the generation of propositions organized at the macrolinguistic level through adequate coherent and cohesive links among the utterances. The macrolinguistic impairments of participants with ASD were likely related to more general difficulties in the prelinguistic conceptual phase of message planning. Namely, they might stem from cognitive difficulties affecting those executive functions that are required to adequately plan a discourse structure, monitor its production, and inhibit the potential production of utterances that are not coherent with the flow of the story (see also Miyake et al., 2000; Mozeiko et al., 2011). Interestingly, in the group of children with ASD both global and local coherence errors negatively correlated with the % of complete sentences suggesting the possibility that an inability to generate a correct mental model of the story induces the production of utterances that are not coherent with the story that, in turn, may frequently trigger a pause in the subsequent phase of grammatical construction. These correlations suggest that the participants' grammatical difficulties were related to their macrolinguistic impairments and support the hypothesis that their difficulties in message planning and organization might have an impact on their grammatical production skills.

According to an influential model (e.g., Levelt, 1989; Levelt et al., 1999), message production is a complex activity that requires different processing stages. The first is a phase of message planning. Here, speakers need to generate the conceptual organization of the message, which includes also the formulation of an internal representation of the addressee's mental model and the use of such representation to subsequently select words with unambiguous referents. This is an area of particular weakness for individuals with ASD. Indeed, even when they can take to some extent the interlocutor-specific prior experience into account, they may produce words whose referent is not always

clear to their listeners (e.g., Arnold et al., 2009; Banney et al., 2015; Malkin et al., 2018). Our data confirm this weakness by showing the presence of a significant amount of a specific type of error of local coherence, i.e., the production of words with ambiguous referents. Therefore, the communicative inefficacy of their narrative samples stems, at least in part, from such inability to use nouns and or pronouns to unambiguously refer to elements of the story. After the phase of message planning, it is necessary to extract lexical concepts from memory and this eventually triggers a phase of lexical selection. At this stage, the identification of the right word can be obtained thanks to the inhibition of potential semantic competitors. As to this regard, the participants with ASD had also lexical retrieval difficulties. Indeed, half of them had clinically significant difficulties in naming (46%) and lexical comprehension (42%). Furthermore, such difficulties were reflected also in a reduced narrative fluency (25% of the participants with ASD had reduced speech rates) and the production of semantic errors (in 59% of the cases). These results suggest that the process of lexical selection is impaired in both modalities (i.e., production and comprehension) in this group of individuals with ASD. Therefore, a difficulty in the process of lexical selection might explain their performance on measures tapping also lexical skills. However, as already observed for their macrolinguistic difficulties, this does not necessarily imply that such difficulties arise from a purely linguistic impairment. Indeed, the ability to select the target lexical item in the mental lexicon requires also additional cognitive skills, such as working memory, attention, and executive functions (e.g., inhibition, monitoring, and planning; Miyake et al., 2000; Mozeiko et al., 2011). For example, the production of semantic paraphasias may stem from a failure in the activation of the right lexical item because of the interferences provided by the semantic competitors. Unfortunately, in this study we did not control for such non-linguistic variables. This is a limitation that should be addressed by future studies.

Notably, according to the model of message production by Levelt et al. after selecting the target word the speaker has access to the information stored in it. During the phase of access to the word's lemma, (s)he becomes aware of the morphosyntactic structure required by the word and will use this information to put the selected item in the right position in the sentence. Furthermore, in the phase of morphological coding (s)he will get access to the morphological information regarding the word. The ASD participants' performance on the sentence completion subtest of the BVL_4-12 and the enhanced production of morphological errors (i.e., % of paragrammatic errors) and omissions of function words in their narrative speech samples suggest that they had also difficulties in the phases of access to morphosyntactic and morphological information. Indeed, the majority of these participants had clinically significant difficulties in such measures: 71% in sentence completion; 67% in syntactic comprehension; 59% in the production of paragrammatic errors; and 51% in the production of complete sentences. These results agree with previous investigations. They highlight the production of utterances characterized by instances of pronoun reversal in ASD (e.g., Kuijper et al., 2017) as well as the presence of persisting morphological difficulties in school-age children with ALI ranging from 6 years up (Roberts et al., 2004; see also Kuijper et al., 2017). Notably, our results suggest that the participants' morphological and morphosyntactic difficulties likely affected their grammatical skills as also shown in previous investigations (e.g., Tuchman et al., 1991; Eigsti et al., 2007). This relation between morphological, morphosyntactic and grammatical difficulties is supported not only by the reduced number of complete sentences on the narrative production task and their impaired performance on the sentence completion and syntactic comprehension tasks. The correlational analyses support such relation. Indeed, their morphological difficulties were related to the production of fewer grammatically well-formed sentences in the narrative production task (see also Condouris et al., 2003; Losh and Capps, 2003).

In conclusion, the results from the current study support the claims about the generalized linguistic difficulties in these children (e.g., Boucher, 2012). As can be seen in **Appendix B**, even if these difficulties are associated to a large within-group variability (e.g., Rapin and Dunn, 2003), 79% of the participants (19 out of 24) performed below normal range in several measures tapping lexical, grammatical and narrative skills, highlighting difficulties at different stages of message planning, organization, and microlinguistic (i.e., lexical and grammatical) processing. Their macrolinguistic impairments were likely related to more general difficulties in the prelinguistic conceptual phase of message planning and mental model generation of the story. Such weaknesses included a difficulty in the non-verbal conceptualization of the story and the generation of an internal representation of the addressee's mental model triggering the production of stories with violations of both local (i.e., production of words with ambiguous referents) and global coherence that significantly contributed to the reduction of the levels of lexical informativeness. Furthermore, the majority of participants with ASD showed also difficulties on tasks assessing lexical selection and grammatical processing skills in both modalities (i.e., production and comprehension). Apparently, only five individuals (21%) showed spared lexical and morphosyntactic skills. However, two of them diverged significantly from the norms in the production of semantic paraphasias, one produced too many paragrammatic errors, and only two did not show any difficulty at the narrative level. Furthermore, only 2 of the 19 participants with ASD with linguistic difficulties did not show any difficulty in comprehension, whereas the rest of the cohort shoed mixed receptive-expressive disorders. Overall, these findings are in line with previous investigations showing similarities in the language impairments observed children with Developmental Language Disorders (e.g., Williams et al., 2008; Bishop et al., 2017) and have both clinical and research implications. From a clinical point of view, they support the efficacy of non-word repetition tasks in detecting the presence of linguistic difficulties in children with ASD (e.g., Kjelgaard and Tager-Flusberg, 2001). They also support the need to adequately assess the linguistic profile of children with ASD by administering not only traditional tasks but also narrative production tasks that allow clinicians to have a clearer picture of the real linguistic skills of persons with ASD and the interconnections between different stages of

message production (e.g., phases of message planning, lexical selection, lexical access, etc. . .). Furthermore, a comprehensive assessment should include also other cognitive skills that may affect narrative processing. For example, in line with previous studies focusing on the potential role of executive functions in narrative discourse (e.g., Miyake et al., 2000; Mozeiko et al., 2011), we speculated that the difficulties in the prelinguistic phase of message planning and conceptual organization might stem from executive functions' difficulties (i.e., impairments in the ability to plan a discourse structure, monitor its production, and inhibit the potential generation of utterances that are not coherent with the flow of the story). However, such measures were not available in the current investigation. Future studies should include also measures assessing executive functions and recruit larger numbers of participants to run regressions models that might allow both researchers and clinicians to explore the proposed causative relation between executive difficulties, macrolinguistic disorganization and microlinguistic impairment in ASD.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available due to confidentiality reasons. Requests to access the datasets should be directed to the corresponding author.

ETHICS STATEMENT

The study received institutional ethics approval by the Ethics' Committee of the Research Institute IRCCS "E. Medea." Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

AM planned the study, ran the statistical analyses, and wrote the manuscript. GV supervised the recruitment of the participants. RM administered the tasks to the children. MO contributed to the analyses. All authors contributed with comments to the interpretation of the results.

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APPENDIX A

An Example of the Narrative Analysis Performed on a Description of the Nest Story

Subject C.S. / Group: ASD / Age: 9 years; 7 months / Gender: Male.

Italian version (original) with errors marked in English:

"Loro^{missingreferent} **vedono il nido di uccelli** / **poi** …*(3 seconds)* / due persone$^{repetitionof2words+repetitionofutterance}$ / due persone$^{repetitionof2words+repetitionofutterance}$ / due persone vedono il nido$^{repetitionof5words+repetitionofutterance}$ / una^{missingreferent} **diceva** / "**puoi prendere il nido** sotto^{paragrammaticerror} " / **allora la** persona^{missingreferent} **prendeva il nido** / Luca^{missingreferent} scappa^{semanticparaphasia} / poi^{filler} **a un certo punto** alcune^{missingreferent} vedeva^{paragrammaticerror} / **dicevano** / si^{missingreferent} **era fatto male** **a+lla gamba** / **poi una** persona^{missingreferent} **era triste** / ^{missingreferent} **è andata in ospedale**…/ una persona era triste$^{repetitionof4words+repetitionofutterance}$ / lui^{missingreferent} **era quasi svenuto** /".

Time 40".

NB In bold the informative words. The complete sentences are underlined.

English translation:

"They see the nest with the birds / then …(3 seconds) / two people / two people / two people see the nest with the birds / one said / "can you take the nest under" / then the person took the nest / Luca runs away *[falls]* / then at some point some seen / said / he had hurt his leg / then a person was sad / went to the hospital / a person was sad / he was almost unconscious /".

Narrative analysis:

Words: 63 / Utterances: 16 / Speech Rate: 95 words per minute / % Semantic Errors: 1.6 % / % Paragrammatic Errors: 3% / % Complete Sentences: 63% / % Local Coherence Errors: 56% / % Global Coherence Errors: 25% / % Lexical Informativeness: 60%.

APPENDIX B

TABLE A1 | Table detailing the performance of each child with ASD on each linguistic measure

ID	1	2	3	4	5	6	7	8	9	10	11
1	−1	−2	0	−1.5	−2	2	2	−2	2	2	−2
2	−2	−1	−2	−2	−1	2	0	0	0	1	−1.5
3	−2	−2	−2	−2	−1	2	0	0	2	2	−2
4	−1.5	−2	0	0	−2	0	2	−2	2	0	0
5	−2	−2	−2	−2	0	0	2	−1.5	2	2	−2
6	−1	−1	0	0	−1	1	0	0	0	2	−1.5
7	−1	0	0	0	−1	0	1,5	0	0	0	0
8	−2	−2	−2	−2	−1	2	0	−2	1.5	0	−1
9	−1.5	−1.5	0	0	0	1.5	1.5	−2	2	0	−1
10	−2	−2	−1.5	−2	−2	0	2	−2	1.5	1.5	−2
11	1.5	0	0	−1.5	−1	2	1.5	−1.5	0	2	−2
12	1	−2	1	−2	0	1	2	0	2	2	−2
13	−2	−2	−2	−2	−2	2	2	−2	2	2	−2
14	0	−2	0	0	2	2	1.5	0	2	0	1.5
15	−2	−2	−2	−2	0	0	2	1.5	1.5	2	−2
16	0	1	1	0	0	1.5	1	0	0	0	1
17	1	−2	1	1.5	1	2	0	−2	2	2	1
18	0	1	0	0	0	2	1	0	1	1.5	1.5
19	0	1	0	1	1	1	0	0	0	0	1.5
20	−2	−2	−2	−2	−2	2	0	−2	2	2	−2
21	−2	−2	−2	1.5	2	2	1.5	−2	2	1.5	1
22	0	1.5	0	1.5	0	0	1.5	0	2	1	1
23	0	−2	−2	−2	1.5	1.5	0	1	0	1	−2
24	1	−2	0	1.5	1	0	0	0	2	0	0

Table showing the z-scores of each participant with ASD on each linguistic measure. Z-scores highlighted in yellow show performances of about 1.5 SDs above (in case of errors) or below expected means. Z-scores highlighted in red show performances of about at least 2 SDs above (in case of errors) or below expected means. Legend: 1. Naming; 2. Sentence Completion; 3. Lexical Comprehension; 4. Grammatical Comprehension; 5. Speech Rate; 6. % Semantic Errors; 7. % Paragrammatic Errors; 8. % Complete Sentences; 9. % Local Coherence Errors; 10. % Global Coherence Errors; 11. % Lexical Informativeness.

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ORIGINAL RESEARCH
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Parental Attunement, Insightfulness, and Acceptance of Child Diagnosis in Parents of Children With Autism: Clinical Implications

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Early parent–child relationships are an important factor influencing many domains of child development, even in the presence of autism. In this study, we investigated the associations between parent–child attunement during play, parental insightfulness, and parental acceptance of their child's diagnosis of an autism spectrum disorder. A sample of 50 parents (26 mothers and 24 fathers) of 26 children aged between 24 and 58 months were videotaped during parent–child play interactions and then interviewed about what they thought had gone through their child's head during the play interaction, and about their feelings and thoughts about their child's diagnosis. Play interactions were evaluated using a coding protocol to assess parental attunement. The results showed that parents who were more able to accept their child's diagnosis and to see things from their child's perspective were more likely to also be attuned during play interactions with their children. These findings highlight the importance of studying the parental ability of insightfulness and acceptance of their child diagnosis of ASD for the implementation of intervention programs for supporting parental attunement and improving the interactions between the parents and the children with autism spectrum disorders.

Keywords: parental attunement, insightfulness, acceptance of child diagnosis, autism spectrum disorders, parent–child interaction

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INTRODUCTION

Autism spectrum disorders (ASDs) are neurodevelopmental disorders characterized by deficits in social communication, and restrictive, repetitive behavioral patterns emerging early in child development. These children also show an intensified emotional reactivity and difficulties in emotion regulation (Samson et al., 2012).

A recent study found a relationship between children's alexithymia and a reduction in parent–child interactions in the presence of a diagnosis of ASD, when compared to parents of typically developed children (Costa et al., 2019). Moreover, a 2019 review have revealed that parental verbal responsiveness to their children's focus of attention predicted children's expressive and receptive language (Edmunds et al., 2019). In this respect, considering that children with ASD may display poorer communicative behaviors than children with typical development, these fewer

social interactions may lead to reduced learning opportunities with parents (Tager-Flusberg, 2016). Given the significance of difficulties in social relationships for these children, many researchers have argued for the need to better understand the role and quality of early relationships with primary caregivers (Crowell et al., 2019).

Research into parent–child dyads highlights the fact that social competence is an important factor in child development (Raver and Zigler, 1997; Vaughan Van Hecke et al., 2007; Denham et al., 2012; Domitrovich et al., 2017). Social competence is shaped within interactive, mutual exchanges as part of the development of early parent–child relationships (Feldman and Masalha, 2010), for which attuned parenting is fundamental (Landry et al., 2006; Leerkes et al., 2009). Parental attunement is a core dimension, defined as the parental ability to be responsive to child signals, understand them, and respond appropriately, while adjusting to the child's needs (Stern et al., 1985; Stern, 1998; Schore, 2001; Zand et al., 2014). This competence emerges during parent–child interactions, laying the foundation for a shared and emotionally connoted experience, which represents a precursor for the development of the child's mind, his/her abilities in self-regulation, and capacity to be engaged in relationships with others.

In research investigating the role of parental attunement in child development, little has involved samples of parents who have children with ASD. A study of 39 parents of children with different diagnoses (including autism) indicated that parents showing greater knowledge of child development were more likely to be attuned to their children. Greater parental attunement also predicted more positive attitudes toward child independence, which in turn predicted child social competencies (Zand et al., 2014). A pilot study examining parent–child physiological synchrony, that is the parent and child electrodermal activity measured during naturalistic free play, highlighted that higher ASD symptoms were associated with lower levels of parental emotional attunement and synchrony (Baker et al., 2015). In a sample of 40 preschoolers with ASD and 40 matched typically developing (TD) peers, children's ability to self-regulate and mother and father parental disciplinary style were explored (Ostfeld-Etzion et al., 2016). The study confirmed what was already emerging in the relevant literature (Feldman and Klein, 2003; Hirschler-Guttenberg et al., 2015), namely, that parents of children with ASDs used the same parental disciplinary style of parents of TD children, and that a more supportive parental disciplinary style was associated with more child self-regulated compliance. According to a 2017 study in an ASD group, mother–child dyadic interactions were more engaged in mismatched emotion-engagement states and children spent more time exclusively with objects than the dyads in the TD group (Guo et al., 2017). Another recent study used a narrative methodology to study fathers' stories of play interaction with their children with ASD aged between 5 and 12 years old. Three narratives emerged from the fathers' stories (action, adjustment, and acceptance), and among them, acceptance narratives were more likely in fathers showing resistance to societal norms of play, acceptance of, and attunement to their children's play interests (Mitchell and Lashewicz, 2018).

An important contribution to understanding the roles played by the parents of children with ASD has been made by Oppenheim and Koren-Karie (Oppenheim et al., 2001; Koren-Karie and Oppenheim, 2018) with the introduction of the concept of parental insightfulness. This refers to the parental ability to see things from the child's point of view. Previous studies have shown that insightful mothers were more sensitive within their interactions with their children, and these mothers are also more likely to have children with secure attachments (Koren-Karie et al., 2002). Furthermore, insightful mothers were found to display higher levels of positive parenting during interactions with their children, regardless of the number of stressful life events experienced by the mother (Martinez-Torteya et al., 2018) when compared to non-insightful mothers. Higher levels of cooperation and co-parenting in triadic interactions when both parents were insightful were also identified, and no differences were found between mothers and fathers in their ability to see things from the child's point of view (Marcu et al., 2016). A recent study (Feniger-Schaal et al., 2019) on 38 mothers of children with intellectual disabilities found that 41% of the mothers showed positive insightfulness and that better capacity for insightfulness was associated with better maternal sensitivity[1] behavior during mother–child interactions when compared to non−insightful mothers.

When comparing a group of clinically depressed vs. non-depressed mothers, Ramsauer et al. (2014) showed lower sensitivity and insightfulness toward their child, in depressed mothers. Based on clinical theorizing, in the presence of a mental illness, parental ability to display attunement/sensitivity and insightfulness toward a child may be somewhat impaired, which may negatively influence parent–child relationship (Oppenheim and Koren-Karie, 2009; Carter and DelCarmen-Wiggins, 2020). However, to our knowledge, there are no studies investigating these variables in a sample of children with ASD.

Within the population of children with ASD, a 2008 study showed that maternal insightfulness did not depend on the severity of ASD or the level of child functioning. Overall, 42% of mothers were found to be insightful and 58% were found to be non-insightful, regardless of the severity of ASD (Oppenheim et al., 2008). Furthermore, in a sample of 39 children with ASD and their mothers, maternal insightfulness and child secure attachment at preschool age predicted better adaptation to developmental tasks, such as school, 4 and 8 years later (Dolev et al., 2014). Consistent with this, a recent systematic review

[1]In this context with sensitivity, we mean "the caregiver's ability to understand and recognize child's signals. In particular, this term refers not only to the parental ability to interpret the emotional and physical states expressed from the early hours of life, but also to the willingness to provide a sufficiently adequate response in terms of timing and contents" (Di Folco et al., 2016, p. 72). We are aware that the debate on attunement is still open. Specifically, the terms *attunement* and *sensitivity*, which have been carefully described within different theoretical paradigms, not without facing some confusion, were proposed with different conceptual terms (Mesman and Emmen, 2013; Di Folco et al., 2016). Maternal sensitivity appeared initially thanks to the observations conducted by Ainsworth (1967) and is generally used within attachment researchers to describe an ability that emerges within the parent–child relationship. However, the term *attunement* was first described by the psychiatrist and psychoanalyst Daniel Stern (Stern et al., 1985) and refers to individual's ability to share affect, empathize, and appropriately respond to another person, not necessarily the child.

on autism and attachment showed that maternal sensitivity and insightfulness support the development of secure attachment in children with ASD (Kahane and El-Tahir, 2015).

As stated by Oppenheim et al. (2009), in cases of the diagnosis of severe pathology, the study of the parental state of mind should include not only insightfulness but also the acceptance or resolution of the diagnosis as "seeing things from the child's point of view must also include understanding and accepting the challenges associated with the child's diagnosis" (Oppenheim et al., 2009, p. 519). Resolution is the process of the integration of this information/emotion [about their child diagnosis] within the parents' representational systems of themselves as parents, of their child, and of the relationship with their child (Pianta and Marvin, 1993, p. 3). Receiving a diagnosis may cause disruption or damage to normal maternal fantasies about a child (Pouillaude, 2018) and negatively interfere with a parent's acceptance of the child's diagnosis and their investment in the child–parent relationship. Lack of acceptance can interfere with the parental ability to integrate the representations of a "healthy" and "ill" child, and with the possibility of focusing their attention on the present and their relationship with the actual child (Marvin and Pianta, 1996; Pianta et al., 1999; Zavattini, 2016).

In studies with children with different diagnoses, the proportion of parents with acceptance of the child's diagnosis varies from 36 to 81% (Lord et al., 2008; Milshtein et al., 2010; Barak-Levy and Atzaba-Poria, 2013; Yirmiya et al., 2015; Dolev et al., 2016; Baiocco et al., 2017). Parental acceptance of the child's diagnosis does not seem to depend on the time passed since receiving the diagnosis (Pianta et al., 1996; Lord et al., 2008; Hutman et al., 2009; Oppenheim et al., 2009; Milshtein et al., 2010; Kearney et al., 2011; Lecciso et al., 2013; Popp et al., 2014), the child's gender (Marvin and Pianta, 1996; Schuengel et al., 2009; Kearney et al., 2011; Yirmiya et al., 2015; Krstić et al., 2016), or parental gender (Lord et al., 2008; Schuengel et al., 2009; Milshtein et al., 2010; Barak-Levy and Atzaba-Poria, 2013; Yirmiya et al., 2015). Instead, it has been found that maternal acceptance of the child's diagnosis relates to more sensitive caregiving during social play (Dolev et al., 2016) and a better maternal perception of their physical health (Reed and Osborne, 2019). Failure in accepting the child diagnosis is linked to higher maternal distress (Lord et al., 2008; Kearney et al., 2011; Krstić et al., 2015), parental depression (Kearney et al., 2011; Krstić et al., 2015), lower levels of emotional support (Sheeran et al., 1997), greater use of avoidance strategies (Freda et al., 2016), and lower maternal sensitivity (Dolev et al., 2016). Few studies have investigated both paternal and maternal acceptance of child diagnosis, but those have found significant gender differences. Mothers low in acceptance, but not fathers, reported more parental negative feelings and more negative impacts of the child's disease on their social life and marriage (Milshtein et al., 2010). Fathers reported higher levels of couple satisfaction if mothers were able to accept their child diagnosis (Sheeran et al., 1997) and mothers were more prone to use an emotional coping style while fathers tended to use a cognitive coping style when they talked about the experience of receiving the diagnosis of the child's illness (Barak-Levy and Atzaba-Poria, 2013).

As for research on the parents of children with ASD, Milshtein et al. (2010) studied 60 fathers and 61 mothers and found that almost 43% were classified as acceptance of the child diagnosis and that for mothers, the acceptance of the diagnosis was associated with a better perception of the child and the impact of raising a child with a disability on family life. Another study (Lecciso et al., 2013) with a sample of 21 mother–child dyads with high-functioning autism showed that accepting mothers of their child diagnosis were better able to see themselves and their children as mental agents, to think of themselves as a secure base, and to not avoid the negative aspects of the relationship. The maternal ability to accept the child diagnosis was associated with the type of diagnosis: in contrast to the results by Milshtein et al. (2010), the researchers found that mothers of children with high-functioning autism were more likely to be accepting of their child diagnosis than mothers of children with Asperger's syndrome.

Seventy-seven parents of recently diagnosed children with ASD were the participants of a study (Poslawsky et al., 2014) that found that parental acceptance of the child diagnosis (also known as Resolution) was associated with less severe autistic symptoms, and demonstrated a substantial stability of the resolution classification relating to the child's diagnosis after 7 months from the first evaluation. Yirmiya et al. (2015) also examined the stability of resolution classification over time (3 years after the first evaluation) among 78 mothers and fathers of children with ASD. At time 2 (3 years after the first evaluation), mothers' acceptance of the child diagnosis was significantly predicted by an increase in maternal anxiety, an increase in the children severity of symptoms, and a longer duration of time since they received the diagnosis. A 2016 paper presented data from a sample of 46 mothers of children with ASD aged between 2 and 8 years, demonstrating that accepting mothers were more likely to be sensitive to their children during play and reported less psychological parental distress and fewer child symptoms compared to mothers low in acceptance (Dolev et al., 2016). A recent study on 84 mothers of children newly diagnosed with ASD showed that mothers low in acceptance had a worsening of maternal health status (in terms of their perception of their symptoms) after 1 year from the time of their child diagnosis, and they perceived to have a poorer health status when compared to mothers more able to accept their child diagnosis (Reed and Osborne, 2019).

Finally, some studies have investigated both parental insightfulness and the acceptance of child diagnosis. A 2009 study of 67 mothers and their children with ASD did not identify a significant association between these two variables, highlighting instead that insightful mothers were more synchronous than non-insightful mothers during play, while mothers able or not able to accept their child's diagnosis did not significantly differ from each other in synchronous behavior during play (Hutman et al., 2009). The maternal ability to accept child diagnosis and maternal insightfulness were both associated with a secure attachment classification in children with ASD (Oppenheim et al., 2009). A further paper also demonstrated that maternal sensitivity mediated the association between

insightfulness/maternal acceptance of the diagnosis and child attachment in a sample of 45 preschool children with ASD (Oppenheim et al., 2012).

The studies discussed above show that, to our knowledge, only one study investigated the relationships between attunement, insightfulness, and acceptance of the child diagnosis, in the presence of a diagnosis of autism for children, focusing exclusively on mothers. The aim of the present study was therefore to examine the relationships between these three aspects of parental functioning, on both mothers and fathers. We hypothesized that the parents of children with ASD who are insightful and able to accept their child diagnosis are more likely to be attuned with their children during play interaction than parents low in their ability to accept their child diagnosis and insightfulness.

MATERIALS AND METHODS

Sample

Participants in this study were 50 parents (24 fathers and 26 mothers) of 26 children who had been diagnosed within the past 3 months of study participation with ASD or being at risk for autism due to a diagnosis of global developmental delay. Children ranged in age from 24 to 58 months (M = 34.36, SD = 8.65) and the total sample comprised 23 (88%) males and 3 (12%) females. Autistic risk was calculated for children under 30 months using the Toddler Module of the Autism Diagnostic Observation Schedule-2 (ADOS-2; Lord et al., 2012a,b). The children with a diagnosis of ASD were 8 (Module 1 Pre-verbal of ADOS-2) and the children with a diagnosis of Global Developmental Delay (GDD) with a risk for autism were 18 (Toddler Module of ADOS-2). Before participating in this study, parents and children have received from 0 to 3 months of intervention. Only one parent refused to participate in the study. The average age of mothers was 38.20 years (SD = 5.51) and the average age of the fathers was 41.38 years (SD = 9.09). Of the parents, 80% were Italian and the remaining 20% were from other countries. Concerning educational level, 14.3% of parents obtained a middle school diploma or lower grade, 46.9% a high school diploma, and 38.8% a university degree or higher (**Table 1**).

Procedure

The parents were recruited at the Institute of -*Blinded for Peer Review*- between 2017 and 2018. Parent–child dyads were videotaped during play interactions lasting 15 min. The play

TABLE 1 | Descriptive statistics.

	***N* (%)**		***M***	**DS**
	Females	**Males**		
Parents' gender	26 (52%)	24 (48%)		
Children's gender	3 (12%)	23 (88%)		
Parents' age			39.755 (Years)	7.570
Children's age			34.360 (Months)	8.651
Length of treatment			1.040 (Months)	1,228
Educational level[#]				
Middle school diploma or lower	7 (14.3%)			
High school diploma	23 (46.9%)			
University degree or higher	19 (38.8%)			
Severity of the symptoms				
Mild	5 (19.2%)			
Moderate	10 (38.5%)			
Severe	11 (42.3%)			
Acceptance of child diagnosis				
Resolved	24 (48%)			
Unresolved	26 (52%)			
Insightfulness				
Insightful	27 (54%)			
Non-insightful	23 (46%)			
Attunement				
Attuned	26 (52%)			
Unattuned	24 (48%)			
Acceptance of diagnosis/Insightfulness				
(A) Resolved/Insightful	21 (42%)			
(B) Unresolved/Non-insightful	20 (40%)			
(C) Unresolved/Insightful or Resolved/Non-insightful	9 (18%)			

[#]*One missing data.*

interactions used for coding the DAOS were the same for assessing parental AI. Parents were then asked to complete a questionnaire and to respond to a videotaped interview lasting about 30–45 min. The clinicians who communicated the diagnosis to the families were different from the team of psychologists in the present study. One of the authors of this study is the clinician who administered the ADOS-2, during the assessment for the diagnosis of autism. No children had received a diagnosis before the assessment at our center. The "at-risk group" was made only by toddlers under 30 months of age and that is why there was no diagnosis of ASD.

Parents were recruited after they have received a diagnosis of ASD or Global Developmental Delay (GDD) with a risk for autism for their children. The child diagnosis was communicated to parents after the diagnostic process carried out at the Institute of Orthophonology (IdO) of Rome (Di Renzo et al., 2015). The Reaction to Diagnosis Interview was administered with regard to the actual diagnosis they had (ASD or GDD with a risk for autism). At the moment of participating in this study, parents and children have received from 0 to 3 months of intervention at our clinical institute. The intervention consisted of 10 h of treatment per week including 6 h of child individual/group therapy, 2 h of school observation and counseling, and 2 h of parental psychological support, carried out by different clinicians than those who conducted the present study (Di Renzo et al., 2020b).

This study was not submitted to an Ethical Committee for ethical review and approval because it is suggested but not mandatory in our legislation. In accordance with articles 5, 7, and 9 of the Italian Ethical Code for Psychologist, a written informed consent to participate in this study was provided by the participants' legal guardian of children. Before participating in the study, parents were asked to sign an informed consent indicating the methods, possible risks, and purpose of the study, as well as being given the possibility of refusing to participate further at any time, in accordance with the Helsinki Declaration (World Medical Association, 2013).

Instruments

The *Autism Diagnostic Observation Schedule*, Second Edition (ADOS-2; Lord et al., 2012a,b; Colombi et al., 2013) is a semi-structured, standardized assessment of communication, social interaction, play, and restricted and repetitive behaviors for children aged between 12 months to adulthood. It presents various activities that elicit behaviors directly related to a diagnosis of ASD. By observing and coding these behaviors, we obtained information relating to two areas: Social Affect (AS) and Restricted and Repetitive Behaviors (RRBs). Critical behaviors in the area of Social Affect, quantified in the coding algorithm, receives a score ranging from 0 to 2, where 0 indicates normotypic behavior, 1 indicates a behavior that is present but atypical and/or not very flexible, and 2 indicates an anomaly or an absence of such behaviors. The RRBs follow a progressive numerical coding based on their frequency and intensity increasing from 0 to 2. The overall score is given by summing AS and RRBs. The ADOS-2 includes five modules: the Toddler Module, for children between 12 and 30 months of age who do not have language or who do not consistently use phrase speech; Module 1, for children from 31 months and older who do not consistently use phrase speech; Module 2: for children of any age who use phrase speech but are not verbally fluent; Module 3, for verbally fluent children and young adolescents; Module 4, for verbally fluent older adolescents and adults. The ADOS-2 has good psychometric properties confirming its usefulness in distinguishing individuals with ASD from other clinical groups (Mazefsky and Oswald, 2006; Gotham et al., 2007, 2009; Lord et al., 2012a,b; Hus and Lord, 2014; Esler et al., 2015).

The *Reaction to Diagnosis Interview* (RDI) (Pianta and Marvin, 1993) is a brief, 15 min interview, aimed at examining parental resolution of the loss/trauma associated with the experience of receiving a child diagnosis of disability or chronic illness. The RDI assesses this acceptance (or lack of acceptance) through videotaping and then coding an individual parent's responses to six standardized questions with specific probes investigating beliefs, memories, and emotional reactions of parents to the news of the child's illness and any changes that have occurred over time. The coding yields the major classifications of Resolved or Unresolved, plus several sub-classifications within each major classification (Pianta and Marvin, 1993). The Resolved parents are those accepting the diagnosis of their child and can describe with balance the changes that may have occurred following the communication of the diagnosis, without continuing to look into the past or to question the possible causes of what happened (Marvin and Pianta, 1996). They show greater acceptance of the situation over time and can describe the difficulties of the disease and the specific characteristics of their child. Unresolved parents provide inconsistent descriptions of the diagnosis experience. They may produce distorted stories, which highlights an inability to describe the reality of the situation, or the story appears confused and it is difficult for the encoder to follow the thread of the discourse. Parents can also experience difficulty in managing their feelings related to the memory of the diagnosis experience and show themselves to be emotionally overwhelmed by anger or pain, or depressed and/or lost in their memories.

The *Insightful Assessment* (IA) (Koren-Karie and Oppenheim, 2004) is a video replay procedure for assessing parental insightfulness. The procedure involves an initial phase in which the parent and the child are videotaped during three different moments of interaction. Then, the parent is invited to watch brief video clips and interviewed regarding his/her child's thoughts and feelings. The evaluation allows each parent to be assigned one of the following categories: Positively Insightful (PI) in which the parent shows that he can describe the child in a complex way and can focus on his internal world; One-sided (Os) in which the parent has a one-dimensional view – positive or negative – of the child and the relationship; Disengaged (De) in which the parent shows a lack of emotional involvement in the description of the child and the relationship; and Mixed (Mx) in which no single, coherent parent strategy emerges (Koren-Karie and Oppenheim, 2018). In our study, parents were divided according to whether they fell into the broad Insightful and Non-insightful classifications (which includes the Os, De, and Mx classifications).

The *Dyadic Attunement Observation Schedule* (DAOS; *under validation*) is an observational measure of parent–child interaction during play. The DAOS observation schedule was used for scoring parent–child dyads videotaped during play

interactions. Parents were invited to play with their children as if they were at home. The clinician gave the parents the instructions of creating three different circumstances lasting about 5–10 min each: a time of free play and two structured playtimes (i.e., blocks and sponge ball). This observational assessment consists of eight scales: 1. joint attention, 2. body, 3. interaction, 4. space sharing, 5. play sharing, 6. authonomy, 7. emotional regulation, 8. understanding child mental states. Each scale has a score ranging from 0 to 3 and the final coding allows parents to be assigned one of two categories: Attuned or Unattuned. Attuned parents can adapt their bodies to respond to their child's signals with combined and alternating use of their space (remaining close to/far, next to/face to face). They are generally able to play with their child by activating a body dialog made up of gestures, sounds, and eye gazing, supporting an interactive exchange in which they organize role switching and involving the child with sufficient participation (without intrusiveness). Attuned parents are also able to facilitate their child's actions without overlap with the child, with the aim of increasing his autonomy and supporting his skills so that the child may experience new actions. They can offer their emotional availability to the child by co-regulating emotions when the child is not able to regulate these by himself. They are also able to recognize and repair moments of failed attunement. Unattuned parents, on the other hand, do not play with their children by activating body dialog (they sometimes look like clumsy or inhibited), and they are not very proactive in involving their child in play. They show little or no shared and alternative use of space, remaining close to/far, next to/face to face), tending to overpower the child or to withdraw following demands for play. These parents may show a strongly passive role, feeling inadequate, and unable to contain and regulate their child's emotions during difficult periods in the interaction. They are powerless to repair moments of failed attunement.

At present, the DAOS has currently been used with children with typical development, learning disabilities, speech disorders, anxiety disorders, and emotion regulation problems. The measure is under validation.

Data Analysis

We used chi-squared tests to examine differences in parental attunement, acceptance of diagnosis, insightfulness, parental gender, parental educational level, and child severity of symptoms. The variable "severity of symptoms" ranging from 1 to 5 (1 = no evidence, 2 = minimum, 3 = mild, 4 = moderate, 5 = severe symptomatology), was created on the basis of the scores from ADOS-2 and clinical observations of the deficits in the quality of communication and relational behaviors calibrated upon children's age.

We used *t*-tests to determine any significant differences between Attuned/Unattuned, Resolved/Unresolved, and Insightful/Non-insightful parents with respect to the variables "children age" and "length of the treatment." In order to investigate our hypothesis that both Resolved and Insightful parents were more attuned with their children with ASD during play interactions, we created a combined variable Resolution/Insightfulness, similar to the approach used by Oppenheim et al. (2009). Three groups of (A) Resolved/Insightful (21), (B) Unresolved/Non-Insightful (20), and (C) Unresolved/Insightful or Resolved/Non-Insightful (9) parents were formed.

A 3 × 2 cross-tabulation was performed to examine differences between this new variable and parental attunement. We used the likelihood ratio (LR) when our data did not meet the assumption of having at least 80% of the cells with an expected count of over 5 for the chi-squared tests.

The differences between the two groups of children with ASD and autistic risk were calculated for the study variables, showing no statistically significant differences between the two groups ($\chi^2 = 1.923$, $p = 0.166$ for attunement; $\chi^2 = 1.087$, $p = 0.297$ for insightfulness; $\chi^2 = 0.855$, $p = 0.355$ for the reaction to diagnosis). We therefore considered the entire sample in further analyses without distinguishing between the two groups.

In line with Rosner (2010), reported by Dogan and Dogan (2015), ICC < 0.4 indicates poor dyadic relationship, so we assumed our dyads had poor relationships for the three main variables of our study (acceptance of diagnosis, insightfulness, and attunement), and we considered mothers and fathers separately for statistical analysis.

RESULTS

Descriptive Analysis

As shown in **Table 1**, 26 parents were classified as Unresolved (15 fathers and 11 mothers); 23 parents were Non-Insightful (16 fathers and 7 mothers), and 24 parents were Unattuned (13 fathers and 11 mothers). Children were assigned to a group according to the severity of symptoms as follows: 3 = mild (19.2%), 4 = moderate (38.5%), and 5 = severe (42.3%). No children were assigned to the groups 1 = no evidence or 2 = minimum.

We examined the associations between insightfulness, acceptance of the diagnosis, and parental attunement with the study variables: parental gender, severity of the child's symptoms, and level of parental education. No differences emerged between mothers and fathers for parental acceptance of the diagnosis ($p = 0.153$) or parental attunement ($p = 0.402$). Significant differences emerged between mothers and fathers relating to insightfulness ($p = 0.005$), with mothers being more insightful than fathers. No significant association was found between the severity of the child's symptoms and RDI classification $p = 0.055$), parental insightfulness ($p = 0.869$), or parental attunement ($p = 0.942$). No significant association emerged between parental educational level and RDI ($p = 0.051$), or parental attunement ($p = 0.145$). The association between parental educational level and parental insightfulness was statistically significant ($p = 0.006$), with insightful parents more likely to have a university degree or higher and non-insightfulness parents more likely to have a high school diploma (**Table 2**).

Furthermore, no significant differences emerged between Resolved/Unresolved ($p = 0.389$), Insightful/Non-insightful ($p = 0.462$), and Attuned/Unattuned ($p = 0.707$) parents

TABLE 2 | Descriptive statistics, associations, and group differences with acceptance of the child diagnosis, insightfulness, and attunement.

	Acceptance of child diagnosis (RDI)		**χ^2**	**Insightfulness (IA)**		**χ^2**	**Attunement (DAOS)**		**χ^2**
	Resolved (% of the total)	**Unresolved (% of the total)**		**Insightful (% of the total)**	**Non-insightful (% of the total)**		**Attuned (% of the total)**	**Unattuned (% of the total)**	
Mothers	15 (30%)	11 (22%)	2.039	19 (38%)	7 (14%)	7.936*	15 (30%)	11 (22%)	0.703
Fathers	9 (18%)	15 (30%)		8 (16%)	16 (32%)		11 (22%)	13 (26%)	
			LR			**LR**			**LR**
Middle school diploma or lower[#]	2 (4%)	5 (10%)	5.950	4 (8%)	3 (6%)	10.360*	3 (6%)	4(8%)	3.859
High school diploma	8 (16%)	15 (31%)		7 (14%)	16 (33%)		9 (18%)	14 (29%)	
University degree or higher	13 (27%)	6 (12%)		15 (31%)	4 (8%)		13 (27%)	6 (12%)	
			LR			**LR**			**LR**
Mild	8 (16%)	2 (4%)	5.803	6 (12%)	4 (8%)	0.283	5 (10%)	5 (10%)	0.120
Moderate	7 (14%)	13 (26%)		10 (20%)	10 (20%)		11 (22%)	9 (18%)	
Severe	9 (18%)	11 (22%)		11(22%)	9 (18%)		10 (20%)	10 (20%)	
	M (DS)	**M (DS)**	***t***	**M (DS)**	**M (DS)**	***t***	**M (DS)**	**M (DS)**	***t***
Children's age	33.250 (7.320)	35.385 (9.753)	0.869	33.518 (7.029)	35.348 (10.316)	0.742	34.808 (7.408)	33.875 (9.966)	0.378
Length of treatment	1.083 (1.380)	1.308 (1.436)	0.678	1.259 (1.534)	1.130 (1.254)	0.247	1.269 (1.538)	1.125 (1.262)	0.219

*$p < 0.01$. [#]*One missing data. LR, Likelihood Ratio.*

according to child age. No significant differences emerged between Resolved/Unresolved (p = 0.501), Insightful/Non-Insightful (p = 0.806), and Attuned/Unattuned (p = 0.828) parents according to the length of the treatment (**Table 2**).

Associations Between Parental Acceptance of the Child Diagnosis/Insightfulness and Attunement

We checked the associations between the variable Resolution/Insightfulness and the study variables that were significantly associated with Insightfulness: level of education and gender of the parents. The association with the educational level was statistically significant (LR = 10.269, df = 4, $p < 0.05$) with Resolved/Insightful parents more likely to have a university degree or higher, and Unresolved/Non-insightful parents more likely to have a high school diploma. The association with the parental gender was not statistically significant (LR = 5.844; df = 2; p = 0.054) for parental gender.

A 3 × 2 Resolution/Insightful × Attuned crosstab (**Table 3**) showed a significant association between the variables (LR = 10.157, df = 2, $p < 0.01$). Parents classified as both Insightful and Resolved were more likely to be Attuned during the play interaction with their children than parents in the other two groups, and parents classified as both Non-Insightful and Unresolved were more likely to be Unattuned during the play interaction with their children than parents in the other two groups.

DISCUSSION

These findings support our hypothesis that parents high in acceptance of their child diagnosis and insightful are more likely to be attuned to children with ASD during play interactions than those low in acceptance and insightful. Parental abilities include understanding their child's point of view and accepting the experience of having received a child's diagnosis of ASD. In addition, being able to focus attention on the present and their relationship with the child, together with the ability to understand the child's perspective taking into consideration his/her mental states, wishes, and difficulties, all appear to be associated with being responsive to a child's signals and responding to these while appropriately adjusting for their needs.

TABLE 3 | Parental acceptance/insightfulness categories and attunement as percentages of the total sample.

	Attuned (% of the total)	Unattuned (% of the total)	LR
(A) Resolved/Insightful	15 (30%)	6 (12%)	
(B) Unresolved/Non-insightful	5 (10%)	15 (30%)	10.157*
(C) Unresolved/Insightful or Resolved/Non-Insightful	6 (12%)	3 (6%)	

*$p < 0.01$.

From our clinical experience, we can assume that the parents able to accept their child diagnosis may better contrast the desires associated to the fantasies about his or her child as "healthy" (Pouillaude, 2018), protecting the child from the projection of unreal desires associated with him/her, or the parent manages to overcome the image of himself as the "parent of an autistic child" and that of the child as an "autistic child," allowing both of them to access a process of individuation and psychic growth. Non-insightful parents are likely to have a rigid and unidimensional (positive or negative) perception of their child's behavior and motivations, may show a lack of emotional involvement or interest providing only short and limited descriptions of the child, or may be very hostile, angry, and concerned about the child (Koren-Karie and Oppenheim, 2018).

Our data lay in the findings from the studies that, within the attachment framework, have shown that in mother–child dyads with the presence of a diagnosis of ASD, the ability to accept and elaborate the experiences of the diagnosis together with the capacity of insightfulness was associated with a secure attachment in children (Oppenheim et al., 2009). Furthermore, the relationship between insightfulness, acceptance of the diagnosis, and child's attachment was mediated by maternal sensitivity (Kahane and El-Tahir, 2015). The acceptance of the child diagnosis along with insightfulness may favor parental ability to be responsive to child signals because they allow the parents to establish a relationship with the "real child," that is, the one whom the parent meets and experiences in terms of strengths and weaknesses and the potential for his/her development. In cases of a severe diagnosis of the child, the parents may become frustrated and disappointed once confronted the "real child" with the "imaginary child," that is the one dreamed of during pregnancy.

Thus, attunement ability can be proposed to promote a secure attachment allowing "the infant to perceive a sense of being accepted and recognized, which facilitates social adjustment and a positive psychological functioning" (Manini et al., 2013). Furthermore, as suggested in the *Introduction*, children's social competence may be positively influenced by attuned parenting, supporting an aspect usually inadequate in children with ASD. This hypothesis should be verified in future research through longitudinal studies. Another interesting finding is that parents low in acceptance or in insightfulness are more likely to be attuned during play interactions with their children, suggesting a possible protective factor of at least one of the two parental abilities to understand child's point of view or to accept the child diagnosis.

The percentage of parents who have accepted the child diagnosis experience and those demonstrating parental insightfulness are consistent with what emerges in other studies with parents of children with ASD (Hutman et al., 2009; Oppenheim et al., 2009; Milshtein et al., 2010; Lecciso et al., 2013; Yirmiya et al., 2015; Dolev et al., 2016). However, some differences to the published findings also emerged in the current study. In our study, the parental acceptance of child diagnosis was not associated with parental gender, child age, or parental educational level. No significant differences emerged for the severity of the children symptoms, in contrast to previous studies

that found that a worsening of the ASD levels of functioning, along with other variables, predicted maternal acceptance of the child diagnosis (Yirmiya et al., 2015) and that the severity of the ASD diagnosis was associated with parental acceptance of the diagnosis (Poslawsky et al., 2014). A possible explanation for these differences concerns the selection of measures that are used to identify the level of severity, such as questionnaires, interviews, or observational tools. The use of different tools can make it difficult to compare the results obtained in different studies. However, we assume that parental acceptance of the diagnosis is associated with parental resilience and previous emotional stability rather than the severity of the child's symptoms, allowing parents to find creative solutions even in the face of serious clinical scenarios.

We found that maternal insightfulness did not depend on the severity of symptoms or child age as ascertained in other studies (Oppenheim et al., 2008). However, our data showed a statistically significant association between parental educational level and parental insightfulness in the direction of higher educational level for insightful parents. This is consistent with findings from the study by Oppenheim et al. (2009) in a sample of mothers of children with ASD, indicating that mothers classified as insightful had a higher level of education than mothers classified as non-insightful. However, this pattern has not been confirmed in samples of mothers of children with typical development patterns (Oppenheim et al., 2001; Koren-Karie et al., 2002), suggesting that this relationship may be specific to samples of children with ASD (Oppenheim et al., 2009). We assume that a higher level of education could function as a protective factor in understanding the child's internal world when it seems that the child deviates from typical functioning, or that a broad cultural background could help parents adapt their resources to the needs of the child. Moreover, a statistically significant association emerged between educational level and the combined variable Resolution/Insightful, with parents both accepting the child diagnosis and insightful more likely to have a university degree or higher, and parents both less able to accept the child diagnosis and less insightful more likely to have a high school diploma.

Finally, significant differences emerged between the insightfulness of mothers and fathers, suggesting that mothers are more insightful. This finding is in contrast with that from another study that tested whether mothers and fathers differ in insightful ability using a low-risk sample of parents of young children with typical development (Marcu et al., 2016). This probably highlights a specific aspect of our sample that should be explored further in future work. It is possible that some fathers may experience greater difficulties than mothers since, in general, they spend less time caring for children (Dyer et al., 2009; Hartley et al., 2014). Some research indicates that many fathers want to increase their levels of involvement in child care if supported on this path (Rankin et al., 2019), which may lead to them feeling frustrated if having less chance of developing an understanding of their child.

This study has several methodological strengths given that narratological and observational measures are less vulnerable to the willingness of participants to provide information or to provide a personal view of the information collected as compared to questionnaires. A further strength is that the literature has often failed to consider the role of fathers whereas we directly tested this. Nonetheless, this study has also some limitations that should be taken into account as they could reduce the generalization of the results. These include the small number of parents who participated, the specific diagnosis of the children involved (risk for autism relating to global developmental delay and autism spectrum disorder), and the use of one measure that is not yet validated in the literature (the Dyadic Parent–Child Attunement Observation Schedule). Literature provides several observational instruments to measure parental attunement, especially within attachment theory researchers. As mentioned above, the debate on attunement is still open and the authors vary in their formulation of this construct (Mesman and Emmen, 2013). The tool we used for the assessment of the parental attunement, which is currently being validated, was specifically built for assessing interaction within parent and child with autism, guided by our theoretical basis and specific therapeutic intervention, focused on children body and sensory processing to promote the ability to be responsive to others' signals, understanding them and replying to them appropriately (Di Renzo, 2017). To overcome these limitations, future studies should involve a larger and less heterogeneous sample and include additional measurements of parental attunement. Furthermore, given the lack of information on the child's level of development and the physical and mental health of the parents in this study, future research should investigate the relationship between these variables and the acceptance of the child's diagnosis, insightfulness, and attunement. Finally, we want to report the cross-sectional design, the use of categorial rather than continuous variables, and the use of parents of the same children as further limitations of our study.

CONCLUSION

The results presented in this study provide some insights into potential clinical work with the mothers and fathers of children with ASD. Studying the parental ability of insightfulness and acceptance of a child diagnosis of ASD has enriched our understanding of the processes underlying the interactions of these parents with their children. These aspects should be addressed through intervention programs for parents. At the Institute of Orthophonology (IdO) support for parents has been incorporated into the D.E.R.B.B.I. intervention (known in full as the Developmental, Emotional Regulation and Body-Based Intervention) within the Turtle Project (Di Renzo et al., 2016). The project combines various interventions offered to children and parents including child assessment (Di Renzo et al., 2019), counseling for parents, clinical sessions with the professionals who work with the child, thematic seminars and experiential workshops, mothers/fathers–child in care settings, and groups of parents (Di Renzo et al., 2020a).

The importance of starting and supporting a process of acceptance relating to the child diagnosis (Guerriero et al., 2017a,b; Guerriero and Di Folco, 2017; Freda et al., 2019; Waizbard-Bartov et al., 2019) and parental

insightfulness could support the relational experiences that determine the child "way of being" that is strongly connected to the non-verbal aspects of parental communication, especially parental attunement (Di Renzo, 2017). According to Trevarthen and Delafield-Butt (2013), responsive and attuned communication and a pattern of timed and sensitive actions can compensate for children experiencing repetition of uncertain and anxious attempts, when psychomotor attunement with perceptive and motor experiences become confused (LaGasse and Hardy, 2013). The basis of this hypothesis is the importance of considering the close interaction between dyadic function and specific parenting abilities in the formation of the psychic structure and the self-regulating abilities of the child (Beebe et al., 1999).

The results of this study also help us to better understand some of the discrepancies between mothers and fathers, which could give useful indications in planning group interventions for parents of different genders.

To date, only a few studies have investigated the needs of parents of children with ASD while paying particular attention to fathers, their involvement in child therapy, and direct involvement in an intervention (Hartley and Schultz, 2015; Rankin et al., 2019). In the present study, we documented that, in our sample at least, mothers are more insightful than fathers, making it understandable that when children show behaviors that are difficult to manage and understand, as in the case of children with ASD, paternal insight may be inadequate. This aspect should, therefore, be considered as the main goal of group therapy aimed at fathers, while monitoring over time the usefulness of such an approach in supporting fathers' ability to "see things from their child's point of view" (Koren-Karie and Oppenheim, 2018, p. 223).

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation, to any qualified researcher.

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

MD and VG conceived the study concept. GZ and VG designed the model and developed the theory. MP and LR recruited the sample and administered the assessments. VG coded the interviews and analyzed the data. FB helped in supervise the project and with MD they provided critical revisions of the findings. All authors discussed the results and contributed to the final manuscript.

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CORRECTION
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Corrigendum: Parental Attunement, Insightfulness, and Acceptance of Child Diagnosis in Parents of Children With Autism: Clinical Implications

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Keywords: parental attunement, insightfulness, acceptance of child diagnosis, autism spectrum disorders, parent–child interaction

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A Corrigendum on

Parental Attunement, Insightfulness, and Acceptance of Child Diagnosis in Parents of Children With Autism: Clinical Implications

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In the original article, there was an error. The acronym "**D.E.R.B.B.I. intervention" was expanded to "Development Emotional Relation Body-Based Intervention**".

An error was also made, referring to "the Institute of -Blinded for Peer Review-"

A correction has been made to ***Conclusion, Paragraph Number 1***:

The results presented in this study provide some insights into potential clinical work with the mothers and fathers of children with ASD. Studying the parental ability of insightfulness and acceptance of a child diagnosis of ASD has enriched our understanding of the processes underlying the interactions of these parents with their children. These aspects should be addressed through intervention programs for parents. At the Institute of Orthophonology (IdO) support for parents has been incorporated into the D.E.R.B.B.I. intervention (known in full as the Developmental, Emotional Regulation and Body-Based Intervention) within the Turtle Project (Di Renzo et al., 2016). The project combines various interventions offered to children and parents including child assessment (Di Renzo et al., 2019), counseling for parents, clinical sessions with the professionals who work with the child, thematic seminars and experiential workshops, mothers/fathers–child in care settings, and groups of parents (Di Renzo et al., 2020a).

The authors apologize for this error and state that this does not change the scientific conclusions of the article in any way. The original article has been updated.

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An Examination of Parent-Reported Facilitators and Barriers to Organized Physical Activity Engagement for Youth With Neurodevelopmental Disorders, Physical, and Medical Conditions

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Organized physical activity (OPA) is an important contributor to physical, social, and emotional health and well-being; however, young people with disabilities are participating at lower rates than their peers without disabilities. This study aimed to (1) compare facilitators and barriers to OPA for young people with disabilities who currently do and do not participate in OPA and (2) to assess whether groups differed in the type of internal and external assets they reported. Parents of 218 young people (41% with a primary diagnosis of autism spectrum disorder) with a diverse representation of disabilities completed an online survey. Young people were categorized as either participants in OPA (n = 131) or non-participants (n = 87) by parent report. Non-participation was significantly predicted by the barrier "there are no activities my child enjoys" and by a lack of children's motivation and happiness during OPA. Significant internal assets differentiating participants from non-participants were the ability to understand simple instructions, love of sport, and meeting physical activity guidelines. Significant external assets were parent and sibling participation in OPA, school type, and household income. The findings from this study have important implications for the design of public health interventions that aim to promote OPA in young people with disabilities, highlighting the need to make activities enjoyable, promote participation of siblings and parents, and support low-income families to participate.

Keywords: organized physical activity, positive youth development, disability, facilitators, barriers

INTRODUCTION

The benefits of regular participation in physical activity (PA) for physical, social, and emotional health and well-being are well-recognized (Janssen and Leblanc, 2010; Ahn and Fedewa, 2011; Moeijes et al., 2018, 2019). One type of PA that children and young people engage in regularly for health and fun is organized physical activity (OPA). OPA is defined as PA organized by a club, association, or other type of organization. It usually comprises training sessions or classes, competitions, or matches supervised or coached by an adult (Australian Sports Commission, 2018; Wiium and Safvenbom, 2019). OPA is an important context in which positive youth development occurs, providing opportunities for social engagement, promoting social skills (Howie et al., 2010), enhancing mental health, and improved quality of life (Cutt et al., 2007; Eime et al., 2013; Moeijes et al., 2019).

Organized physical activity has the potential to provide additional benefits for young people with disability such as promoting inclusion, providing social connection, reducing complications of immobility, enhancing social and emotional well-being, and controlling or slowing functional decline (Murphy and Carbone, 2008; Rosewater, 2009; Anderson and Heyne, 2010; Howells et al., 2019). Yet, despite the plethora of benefits, children and young people with a disability are less likely to engage in OPA than those without a disability (Solish et al., 2010; Shields and Synnot, 2016). To understand the low rates of participation, studies have examined facilitators and barriers to PA in children and young people with a disability (Shields et al., 2012; Martin, 2013); however, there is little research focused explicitly on OPA. This is an important gap as there is evidence to suggest that different personal and environmental factors are associated with OPA participation compared to unorganized (or self-organized) PA (Smith et al., 2010; Noonan et al., 2017; Wiium and Safvenbom, 2019). Furthermore, participation rates are lower for OPA than for unorganized PA in children with a disability (Solish et al., 2010; Arim et al., 2012).

Several theoretical frameworks have been used to understand PA participation of children with disabilities (Ross et al., 2016). They include the theory of Planned Behavior (Ajzen, 1991), self-determination theory (Deci and Ryan, 2000), and socio-ecological frameworks (e.g., Bronfenbrenner, 1979; see Rhodes et al., 2019 for a review of these theoretical frameworks). To address conceptual and terminological inconsistencies relating to the participation construct, the Family of Participation-Related Constructs (fPRC) framework was proposed (Imms et al., 2016, 2017). Participation was considered to include attendance and involvement, the latter being viewed as the experience of participation during attendance, which encompassed motivation, persistence, engagement, social connection, and affect (Imms et al., 2016). Three concepts related to participation and incorporated into the framework were preferences (activities that hold meaning or are valued), sense of self (confidence, satisfaction, self-esteem, and self-determination), and activity competence (capability capacity, performance). The relationships between these within-person factors and participation were hypothesized to be bi-directional; influenced by past participation experiences and, in turn, influencing future participation.

While no theory or combination of theories has sufficiently explained all the variables associated with PA (Bauman et al., 2002), the integration of components from various theories into a multilevel framework is thought to offer the best way to understand and intervene in PA behavior (King et al., 2009; Bauman et al., 2012). One such framework which integrates multiple theories across multiple levels is the positive youth developmental (PYD) framework, a strengths-based interdisciplinary approach that links the young person's developmental strengths and inherent capacity to thrive with ecological contexts (relationships, resources, communities, opportunities) (Benson et al., 2007). In the PYD framework, OPA is considered to be a developmental context in which PYD can be promoted (Zarrett et al., 2008). It is noted in the PYD literature, however, that OPA does not automatically result in positive outcomes. Aligning with the fPRC framework which distinguishes between attendance and involvement, proponents of PYD suggest that positive outcomes are contingent on the way that OPA is delivered and experienced by the young person (Petitpas et al., 2005). Specifically, the activity needs to be intrinsically rewarding, provide opportunities to learn or acquire life skills (internal assets) such as problem solving and decision-making, and be supported by external assets such as caring adult mentors (coaches), strong peer relationships, parental involvement, and a sense of belonging to the wider community (Holt et al., 2017). The PYD framework fits well within the disability context because of its emphasis on strengths rather than deficit; it positions the young person with a disability as having the same inherent potential to grow and develop as any other young person if they receive support, empowerment, and engagement through positive relationships, contexts, and ecologies (Benson et al., 2007).

While OPA participation is recognized as an important context in which to promote PYD (Holt et al., 2017), many barriers were identified for young people with disabilities in a systematic review of facilitators and barriers to participation (Shields et al., 2012). Personal barriers included lack of skill or coordination, preference for other activities, fear of injury, fear of being teased, not knowing what to do, self-consciousness, previous bad experiences, lack of time, and pain or discomfort. Social barriers included parental attitudes and behavior (e.g., lack of support, time, money, and opportunity), lack of friends, and negative attitudes of others. Environmental barriers included inadequate and inaccessible facilities and lack of transport. Policy and program barriers included lack of appropriate activities, lack of trained staff, negative attitudes of staff, and cost (Shields et al., 2012). While many of the facilitators and barriers were common to those identified in research involving children and young people without disabilities, for example, a child's preference for the activity, cost, and time constraints, others were more clearly related to their disability; these included pain or discomfort, fear of incontinence, fear of being teased (Kang et al., 2007), negative perceptions of disability (from peers, staff, and others), and inadequate or inaccessible facilities and/or programs. The review did not examine, however, whether facilitators and barriers

differed between young people engaged in OPA and those who did not participate or examine whether the type of disability and level of support needs influenced participation in OPA, as has been documented in prior studies (Mâsse et al., 2012; Darcy et al., 2016).

This is the first study, to our knowledge, that compared facilitators and barriers to OPA in two groups of young people with disability: those who currently participated in OPA and those who did not participate. This is an important distinction as, arguably, barriers and facilitators endorsed more frequently by non-participants would be an important focus for interventions to improve participation. This study also focused explicitly on OPA, rather than the broader topic of PA, due to research indicating that participation rates are lower for OPA than for PA for young people with disability (Solish et al., 2010; Arim et al., 2012). Situating the study within a PYD framework, this study addressed two research questions: (1) Do parents of young people with disability who do not participate in OPA differ in the facilitators and barriers they perceive compared to those who do participate in OPA? (2) Do young people with disability who do not participate in OPA differ in the type of individual assets (disability type, level of support needed, strengths, regular PA) and external assets (supportive relationships, communities, opportunities, financial resources) to those who do participate in OPA?

Potential facilitators were drawn from the fPRC, namely, the within-person factors (preferences, activity competence, and sense of self) hypothesized to be associated with participation and the young person's involvement (motivation, persistence, social connection, happiness) during OPA. Potential barriers were identified from a review of the OPA literature (King et al., 2009; Shields et al., 2012). Participation was hypothesized to be positively associated with OPA that was experienced as intrinsically rewarding and offered opportunities to learn and acquire life skills such as persistence (internal assets) and to be positively associated with young people who had supportive relationships, resources, and opportunities (external assets). Supportive relationships were operationalized in this study as sibling and parental involvement in OPA, and positive coaching style. Resources and opportunities were measured by household income, access to OPA (distance, cost, time, environment), and National Disability Insurance Scheme (NDIS) support. In Australia, individuals with permanent and significant disability can apply to be supported by the NDIS which provides funding for supports and services. Families in this study were asked whether their young person was supported by the NDIS which was posited as an external asset as these supports can be used to assist with daily living, to participate in community activities, to increase independence, and to pursue goals. Other potential barriers and facilitators were measured by comparing participants and non-participants on demographic factors, parent-reported child strengths, and parent-reported amount and frequency of moderate and vigorous PA. In line with the abovementioned research questions, this study aimed to (1) compare facilitators and barriers to OPA for young people with disabilities who participated and those who did not participate and (2) utilize a PYD framework to assess whether the groups differed in the type of internal and external assets they reported.

MATERIALS AND METHODS

Participants

The sample comprised 218 young people aged 4–17 years (mean age: 10.58) with a diverse representation of disabilities. They were categorized as being current OPA participants (n = 131) or not participating in OPA (n = 87) after completion of an online survey by parents or guardians. Surveys were completed by parents/guardians in this study to avoid over-burdening children and youth with the comprehensive list of barriers and facilitators we were interested in. Ninety-four percent of parent/guardians who responded were female (mean age: 42.9). When young people had more than one condition associated with disability, the condition with the greatest impact (as identified by the parent) was designated as the primary condition. **Table 1** presents the frequency of primary disability categories, number of comorbidities, level of support, and their association with participation in OPA.

Procedure

The study was approved by the Deakin University Human Research Ethics Committee (2016-336). An Australia-wide purposive sampling strategy was conducted by advertising through the Australian National Disability Insurance Agency (NDIA) portal, the Australian Football League (AFL) and various sporting clubs, disability support organizations and Facebook pages. The advertising material consisted of a promotional flyer with a link to an online survey, Plain Language Statement, and consent information. Organizations were asked to promote the advertising material through their appropriate channels (e.g., websites and E-newsletters). Hardcopy advertising materials were also available for the organizations that wished to distribute them, for example, in clinic waiting rooms. Participants most frequently reported hearing about the survey via online social media (n = 91), followed by a disability support organization (n = 57). Fewer participants heard about the survey from "other" sources (n = 26), a sporting club (n = 14), word-of-mouth (n = 10), and the AFL (n = 4).

Materials

The online survey was administered using the survey platform Qualtrics with items developed by a team of health professionals including pediatricians, psychologists, physiotherapists, sports scientists, and public health experts. It consisted of 99 items encompassing child and parent demographic questions, questions pertaining to the young person's disability, current OPA participation, level of moderate and vigorous PA, and a list of facilitators and barriers to OPA (see Supplementary Materials for a copy of the survey).

Twelve potential barriers identified from a review of the OPA literature (King et al., 2009; Shields et al., 2012) were listed with a five-point Likert scale (strongly disagree, disagree, not sure, agree, strongly agree). They included individual barriers

TABLE 1 | Primary condition, comorbidities, level of support, and OPA participation.

Primary condition	Total frequency (%)	Current OPA *N* = 131 (60.09)	No OPA *N* = 87 (39.91)	*p*
ASD	89 (40.83)	51 (57.30)	38 (42.70)	0.381
Cerebral palsy	20 (8.77)	12 (60)	8 (40)	
Intellectual disability	19 (8.72)	13 (68.42)	6 (31.58)	
Down syndrome	14 (6.42)	9 (64.29)	5 (35.71)	
Depression/anxiety	15 (6.58)	6 (40)	9 (60)	
Attention deficit hyperactivity disorder	15 (6.58)	10 (66.67)	5 (33.33)	
Vision impairment	10 (4.59)	5 (50)	5 (50)	
Hearing impairment	8 (3.51)	8 (100)	0 (0)	
Rare genetic	7 (3.07)	4 (57.14)	3 (42.86)	
Diabetes	4 (1.75)	4 (100)	0 (0)	
Epilepsy	5 (2.19)	3 (60)	2 (40)	
Severe speech disorder	5 (2.19)	3 (60)	2 (40)	
Spina bifida	4 (1.75)	1 (25)	3 (75)	
Developmental coordination disorder	2 (0.88)	1 (50)	1(50)	
Other	1 (0.44)	1 (100)	0 (0)	
No. comorbidities				0.952
None	67 (30.73)	42 (62.69)	25 (37.31)	
One	46 (21.10)	28 (60.87)	18 (39.13)	
Two	42 (19.27)	23 (54.76)	19 (45.24)	
Three	30 (13.76)	19 (63.33)	11 (36.67)	
Four	18 (8.26)	11 (61.11)	7 (38.89)	
Five or more	15 (6.88)	8 (53.33)	7 (46.67)	
School support				0.308
None	58 (27.49)	31 (53.45)	27 (46.55)	
2–3 days per week	72 (34.12)	48 (66.67)	24 (33.33)	
3–5 days per week	81 (38.39)	49 (60.49)	32 (39.51)	
Ability to walk unassisted				0.253
Yes	201 (92.20)	123 (61.19)	78 (38.81)	
No	17 (7.80)	8 (47.06)	9 (52.94)	
Ability to understand simple instructions				0.003
Yes	202 (92.66)	127 (62.87)	75 (37.13)	
No	16 (7.34)	4 (25)	12 (75)	

(child preferences, lack of skill, fear of injury, social difficulties), family barriers (time, cost), and environmental barriers (distance, cost, coaching style, unsuitable environment, activities too challenging, or not challenging enough). Items were recoded from "strongly disagree," "disagree," and "not sure" to a binary variable: 0 = no barrier. "Agree" and "strongly agree" were recoded to a binary variable: 1 = barrier.

Using the fPRC model of participation-related constructs, five factors hypothesized to contribute to the young person's involvement in OPA (motivation, persistence, social connection, happiness, and involvement in the activity) were presented with a five-point Likert scale. Items were recoded from "does not describe my child" and "describes my child slightly well" to a binary variable: 0. Items were recoded from "describes my child moderately well," "describes my child very well," "describes my child extremely well" to a binary variable: 1. Three facilitators relating to the young persons' preference for OPA (importance, meaningfulness, preference), two related to activity competence (improvement in skill and performance, increased level of independence performing the activity), and three facilitators relating to sense of self (confidence in ability to perform the activity, general self-confidence, and feelings of satisfaction and pride) were presented with a five-point Likert scale and recoded to a binary variable. If the young person was not currently involved in OPA, parents were asked to respond based on past involvement in OPA.

Parents were asked to report their child's level of moderate and vigorous PA, any positive or negative experiences of OPA, and whether any siblings participated in OPA. The level of support needed by the young person was measured with three items ("does your child receive additional support in school," "is your child able to walk without assistance," and "is your child able to understand simple instructions"). Utilizing a strengths-based approach, parents were asked to list their child's strengths which were then categorized using the Values in Action (VIA) classification of strengths (Wagner et al., 2019).

Analysis Plan

The five-point Likert scale responses were recoded into binary variables. Although this method can diminish power, the

relationship between the underlying construct (perception of barrier vs. non-barrier) and the dependent variable (OPA participation) was not necessarily linear; hence, a binary split was considered appropriate. Chi-square tests were conducted to explore if OPA participants and non-participants differed in their disability condition, number of comorbidities, the facilitators and barriers to OPA they endorsed, or their demographic characteristics. Factors were included in binary logistic regressions based on theory for facilitators (the inclusion of constructs from the fPRC model) and significance value for barriers (barriers that were significant at $p < 0.05$ in the chi-square analyses) to identify significant predictors of participation. Three separate binary logistic regressions were undertaken to identify significant barriers and facilitators (research question 1) and significant internal/external assets (research question 2).

RESULTS

Sixty percent of the sample ($n = 131$) were currently engaged in OPA which is less than the estimated 74% participation rate of OPA for all Australian children (Australian Sports Commission, 2018). The most common activities engaged in by participants in this study were swimming, soccer, dance, basketball, and gymnastics which is identical to the top six activities for all Australian children in 2017 (Australian Sports Commission, 2018). In this sample, 24% of the participants who currently engaged in OPA participated four or more times a week, 53% participated two to three times a week and 23% participated once a week. Parents were asked to report the amount of time their child spent in moderate and/or vigorous PA per week. This data was used to calculate whether the young people in the study were meeting the Australian government's PA guidelines of 60 min of moderate to vigorous PA per day. Only 35% of the sample met this PA guideline which is similar to rates for Australian children in general (30% of children aged 2–17 met the PA guidelines; Australian Institute of Health and Welfare, 2018). Frequency of OPA participation was significantly associated with meeting PA guidelines [$\chi^2(3) = 26.27$, $p = 0.000$, Cramer's V = 0.355]. Participants who engaged in OPA four or more times a week were almost 10 times more likely to be meeting PA guidelines compared to young people who did not participate at all, $b = 2.29$, $p = 0.000$, OR = 9.86, 95% CI [3.79, 25.64]. Participants who participated two to three times were three times more likely to be meeting PA guidelines than non-OPA participants, $b = 1.11$, $p = 0.003$, OR = 3.04, 95% CI [1.45, 6.34].

Although there was a wide representation of disabilities in the sample, 41% listed autism spectrum disorder (ASD) as their primary diagnosis. Sixty-nine percent of the sample had at least one comorbid condition, and 48% of the sample had two or more conditions. There was no significant difference in OPA participation between disability types or the number of comorbidities. Young people who could not understand simple instructions were significantly less likely to be OPA participants. **Table 2** presents demographic information for young people with disabilities and their families. The only significant difference between those who participated in OPA and those who did not was household income and type of schooling. Those families in the lowest income bracket [$\chi^2(2) = 6.80$, $p = 0.033$, $\phi = 0.208$] and young people not attending mainstream school were significantly less likely to be engaged in OPA [$\chi^2(1) = 5.04$, $p = 0.025$, $\phi = 0.149$].

Research Question 1: Do Parents of Young People With Disability Who Do Not Participate in OPA Differ in the Facilitators and Barriers They Perceive Compared to Those Who Do Participate in OPA?

Table 3 presents the percentage of barriers endorsed by parents of OPA participants and non-participants and chi-square analyses. Six barriers were endorsed significantly more by parents of children *not* currently participating in OPA. Using these six barriers as predictor variables in a binary logistic regression (see **Table 4**), the model explained 21% of the variance in OPA participation [$\chi^2(6) = 26.39$, $p = 0.000$, $R^2 = 0.21$]. The only predictor that remained significant in the model was "there are no activities available that my child enjoys." Young people of parents who endorsed this barrier were almost four times as likely to *not* be participating in OPA.

Table 5 presents the percentage of facilitators endorsed by parents of OPA participants and non-participants and chi-square analyses. Using the facilitators in a binary logistic equation (see **Table 6**), the model explained 32% of the variance in OPA participation [$\chi^2(13) = 38.84$, $p = 0.000$, $R^2 = 0.32$]. The only predictors that remained significant in the model were motivation and happiness. Young people of parents who reported that their child appeared unmotivated during OPA were 20 times more likely to *not* be participating in OPA and those that were unhappy during the activity were 12 times more likely to *not* be participating.

Research Question 2: Do Young People With Disability Who Do Not Participate in OPA Differ in the Type of Individual Assets (Disability Type, Level of Support Needed, Strengths, Regular PA) and External Assets (Supportive Relationships, Communities, Opportunities, Financial Resources) to Those Who Do Participate in OPA?

Individual Assets

Organized physical activity participation was not significantly associated with disability type, number of comorbidities, or ability to walk unassisted (see **Table 1**). Three individual factors were associated with OPA participation: ability to understand simple instructions, regular PA, and enjoyment of sport and/or PA. Young people who were not able to understand simple instructions were less likely to be OPA participants [$\chi^2(1) = 8.87$, $p = 0.003$, $\phi = 0.202$]. Young people meeting recommendations of 60 min of moderate to vigorous PA per day were more likely to be OPA participants [$\chi^2(1) = 18.02$, $p = 0.000$, $\phi = 0.289$] and young people who enjoyed sport and/or PA (listed as a strength by their

TABLE 2 | Demographic characteristics of the sample.

		OPA (%)	Not in OPA (%)	*p*
		***N* = 131 (60.09)**	***N* = 87 (39.91)**	
Gender				0.055
	Male	75 (55.15)	61 (44.85)	
	Female	56 (68.29)	26 (31.71)	
Age in years				0.496
	4–8	38 (53.52)	33 (46.48)	
	9–11	40 (60.61)	26 (39.39)	
	12–14	27 (67.50)	13 (32.50)	
	15 +	26 (63.41)	15 (36.59)	
Indigenous status		4 (57.14)	3 (42.86)	0.838
Main language not English		0	3 (100)	0.059
Family type				0.581
	Single parent	18 (52.94)	16 (47.06)	
	Both parents	87 (62.59)	52 (37.41)	
	Other family	10 (58.82)	7 (41.18)	
	Missing	16 (57.14)	12 (42.86)	
Employment status of parent				0.081
	Full-time	31 (65.96)	16 (34.04)	
	Part-time	56 (63.64)	32 (36.36)	
	Home duties	20 (45.45)	24 (54.55)	
	Student/volunteer	8 (80)	2 (20)	
	Missing	16 (55.17)	13 (44.83)	
Household income				0.033*
	Below median HI	19 (46.34)	22 (53.66)	
	Middle median HI	39 (67.24)	19 (32.76)	
	Above median HI	41 (70.69)	17 (29.31)	
	Missing	32 (52.46)	29 (47.54)	
Parent education				0.819
	Year 10 or equivalent	14 (53.85)	12 (46.15)	
	Year 12 or equivalent	41 (61.19)	26 (38.81)	
	Certificate/diploma	35 (64.81)	19 (35.19)	
	Bachelor degree	25 (59.52)	17 (40.48)	
	Missing	16 (55.17)	13 (44.83)	
No. of siblings				0.470
	None	17 (54.84)	14 (45.16)	
	One	53 (67.09)	26 (32.91)	
	Two	29 (59.18)	20 (40.82)	
	Three +	16 (53.33)	14 (46.67)	
	Missing	16 (55.17)	13 (44.83)	
Education				0.036*
	Mainstream	103 (64.78)	56 (35.22)	
	Special	24 (51.06)	23 (48.94)	
	Other	4 (33.33)	8 (66.67)	
NDIS supported				0.814
	Yes	35 (61.40)	22 (38.60)	
	No	96 (59.63)	65 (40.37)	

* *Indicates* $p < 0.05$.

parent) were more likely to be OPA participants [$\chi^2(1) = 4.54$, $p = 0.033$, $\phi = 0.144$].

Supportive Relationships, Communities (External Assets)

Two factors were significantly associated with greater OPA participation: parent involvement in OPA (the parent volunteers as a coach) [$\chi^2(1) = 4.59$, $p = 0.032$, $\phi = 0.145$] and having a sibling participating in OPA [$\chi^2(1) = 12.57$, $p = 0.000$, $\phi = 0.249$]. Coaching style was not significantly associated with participation [$\chi^2(1) = 0.51$, $p = 0.477$].

Resources and Opportunities (External Assets)

Two factors differed significantly between OPA participants and non-participants. Those families in the lowest income bracket [$\chi^2(2) = 6.80$, $p = 0.033$, $\phi = 0.208$] and young people

TABLE 3 | Frequencies and Chi-square results for barriers to OPA.

Barriers	Current OPA		No OPA			
	N	%	*n*	%	$\chi^2(1)$	*p*
No activities my child enjoys	12	9.83	25	33.78	17.25	0.000
Too far to travel	34	27.87	32	43.24	4.86	0.027
Unsuitable/inconvenient time	26	23.21	26	36.62	3.84	0.050
Activities too costly	68	56.67	39	53.43	0.19	0.660
Do not have time to attend	26	22.03	11	15.49	1.21	0.272
Difficulty performing activities	58	52.25	43	74.14	7.59	0.006
Environment not suitable	27	24.32	23	40.35	4.63	0.031
Worries about being hurt/injured	24	20.17	24	32.88	3.90	0.048
Difficulty socially with peers	66	55.00	48	65.75	2.17	0.141
Activities too challenging	51	42.86	43	58.90	4.66	0.031
Activities not challenging enough	5	4.35	3	4.17	0.00	0.952
Coaching style not suitable	31	28.81	21	33.33	0.51	0.477

TABLE 4 | Binary logistic regression for associations of barriers to OPA participation.

Barriers	*B*	*SE*	Significance	OR	95% CI
No activities my child enjoys	−1.39	0.46	0.002	0.25	0.10, 0.61
Too far to travel	−0.71	0.39	0.071	0.49	0.23, 1.06
Difficulty performing activities	−0.21	0.44	0.648	0.82	0.34, 1.97
Environment unsuitable	−0.51	0.42	0.231	0.60	0.26, 1.38
Worries about being hurt/injured	−0.23	0.44	0.609	0.80	0.33, 1.90
Finds activities too challenging	−0.45	0.42	0.287	0.64	0.28, 1.46
Constant	1.80	0.36	0.000	6.02	

TABLE 5 | Frequencies and Chi-square results for facilitators to OPA.

Facilitators	OPA		No OPA			
	n	%	*n*	%	$\chi^2(1)$	*p*
Preference for OPA						
Activity important to young person	100	81.30	28	63.64	5.65	0.017
Activity meaningful to young person	98	80.99	27	61.36	6.77	0.009
Preference for the activity	80	66.67	22	50.00	3.80	0.051
Involvement in OPA						
Appears motivated during the activity	108	87.80	21	47.73	29.62	0.000
Persists throughout the activity	96	78.05	24	53.81	7.86	0.005
Feels a social connection	89	72.36	21	48.84	7.89	0.005
Appears to be happy	106	86.18	32	74.42	3.14	0.076
Appears involved in the activity	105	85.37	27	61.36	11.27	0.001
Activity competence/sense of self						
Increase in skill and performance	110	90.16	31	70.45	9.82	0.002
Increased independence performing activity	113	91.87	29	65.91	17.16	0.000
Confidence in ability to perform activity	111	90.24	28	63.64	16.44	0.000
General self-confidence	105	85.37	29	65.91	7.74	0.005
Feelings of satisfaction and pride	108	87.81	31	70.45	6.99	0.008

not attending mainstream school were significantly less likely to be engaged in OPA [$\chi^2(1) = 5.04$, $p = 0.025$, $\phi = 0.149$]. None of the other environmental factors examined in the study (cost, OPA environment and accessibility, distance to travel, coaching style, the competitiveness of other children and parents, NDIS support) were significantly different between the two groups. Using the three individual and four external assets in a binary logistic regression (**Table 7**), the young person's love of sport, meeting PA recommendations and household income were significantly associated with

TABLE 6 | Binary logistic regression for associations of facilitators to OPA participation.

Facilitators	*B*	*SE*	Significance	OR	95% CI	χ^2	Significance	R^2
Step 1 Preference for activity								
Importance of activity	−0.59	0.89	0.508	0.56	0.10, 3.16	5.59	0.134	0.05
Meaningfulness of activity	0.85	0.82	0.298	2.35	0.47, 11.75			
Preference for activity	0.22	0.52	0.676	1.24	0.45, 3.44			
Step 2 Involvement in OPA								
Motivation during activity	3.03	0.86	0.000	20.59	3.84, 110.46	27.14	0.000	0.27
Persistence throughout activity	−0.92	0.77	0.233	0.40	0.09, 1.81			
Feels a social connection	0.42	0.53	0.420	1.53	0.56, 4.28			
Appears to be happy	−2.57	1.01	0.011	0.08	0.01, 0.56			
Appears involved in activity	0.83	0.92	0.365	2.30	0.38, 13.98			
Step 3 Activity competence/sense of self								
Increase in skill and performance	0.15	1.04	0.883	1.17	0.15, 8.98	6.12	0.295	0.32
Increased independence performing activity	1.06	1.17	0.365	2.88	0.29, 28.39			
Confidence in ability to perform activity	0.92	0.90	0.311	2.50	0.43, 14.64			
General self-confidence	−0.27	1.05	0.796	0.76	0.10, 5.98			
Feelings of satisfaction and pride	−0.67	1.07	0.532	0.51	0.06, 4.18			

TABLE 7 | Binary logistic regression for associations of internal and external assets to OPA participation.

Internal and external assets	*B*	*SE*	Significance	OR	95% CI
Understands simple instructions	−0.87	0.91	0.339	0.42	0.07, 2.51
Meets PA recommendations	1.33	0.46	0.004	3.79	1.54, 9.33
Enjoys sport	1.31	0.58	0.023	3.71	1.20, 11.52
Parent participates in OPA	1.10	1.11	0.321	3.01	0.34, 26.65
Sibling participates in OPA	−0.44	0.43	0.305	0.64	0.28, 1.50
Household income	0.66	0.26	0.011	1.93	1.17, 3.19
School type	−0.46	0.34	0.171	0.63	0.32, 1.22
Constant	0.58	1.26	0.648	1.78	

current OPA participation [$\chi^2(7) = 30.88$, $p = 0.000$, $R^2 = 0.26$].

DISCUSSION

This study aimed to compare facilitators and barriers to OPA for young people with disabilities who participated in OPA and those who did not participate and, utilizing a PYD framework, assess whether the groups differed in the type of internal and external assets they reported. Non-participation in OPA was significantly predicted by the barrier "there are no activities my child enjoys" and by a lack of motivation and happiness during OPA. Significant internal assets differentiating participants from non-participants were the ability to understand simple instructions, the parent-reported strength "love of sport/physical activity," and meeting PA recommendations. Significant external assets were parent and sibling participation in OPA, school type (mainstream education), and household income.

In this study, motivation was the greatest predictor of participation. Parents who reported that their child was unmotivated when they participated in OPA (either currently or during past participation) were almost 20 times less likely to be currently participating in OPA. This finding accords with prior research findings that motivation is an important determinant of PA participation in both children and adults (Hurkmans et al., 2010; Pannekoek et al., 2013). For children, it is primarily intrinsic motivation, derived from the enjoyment of the PA itself, that is associated with participation in PA (Saebu and Sørensen, 2011; Sebire et al., 2013). This accords with the PYD position that activities need to be intrinsically rewarding if positive growth is to occur (Petitpas et al., 2005). Young people in this study who expressed happiness during OPA were 12 times more likely to be current OPA participants. Conversely, young people of parents who endorsed the barrier "there are no activities available that my child enjoys" were significantly less likely to be current participants. This is consistent with research indicating that continuous participation in OPA was contingent upon the enjoyment of the activity in studies of young people with disabilities (Heah et al., 2007; Nyquist et al., 2016) and those without disability (Garn and Cothran, 2006).

The importance of supportive relationships, resources, communities, and opportunities for positive youth development through OPA (Benson et al., 2007) was assessed in this study by examining the environment in which the activity occurs (suitability, distance to travel, level of competitiveness), the relationships (parental involvement, coaching style, peer interactions, sibling participation), and resources (household

income, cost, availability of suitable programs). The only factors that differed significantly between participants and non-participants were school type, sibling and parent involvement in OPA, and household income. Families in the lowest-income bracket were five times more likely to be non-participants in this study, which suggests that costs associated with OPA were a significant barrier. In a large Australia-wide study of children's participation in OPA led by the Australian Sports Commission, only 58% of children from low-income families participated in OPA compared to 84% from high-income families (AusPlay; Australian Sports Commission, 2018). Similarly, international studies have found that young people from lower socioeconomic status (SES) households are engaged in less OPA programs (Sallis et al., 1996; Kantomaa et al., 2007; Brockman et al., 2009). Interestingly, there is evidence to suggest that SES can also influence the type of support that parents provide to facilitate PA. Although this was not examined in the present study, previous research has found that higher SES families were more likely to enroll their children in a variety of OPA and co-participate in activities, whereas lower SES families were more likely to offer verbal encouragement and have children engaged in unstructured activities including outdoor play (Brockman et al., 2009; Noonan et al., 2017). While cost is a barrier that affects families with and without a child with disability, the cost of participation may be particularly onerous for families caring for a child with a disability due to the additional costs associated with disability care (therapies, equipment, loss of earnings due to parental care commitments) (Shields and Synnot, 2016).

Supportive relationships were assessed by examining parental involvement, coaching style, peer interactions, and sibling participation. The only factors associated with participation were sibling and parental involvement in OPA. Parents are recognized as one of the most important influences of PA in their children (Beets et al., 2010; Edwardson and Gorely, 2010; Smith et al., 2010), and many studies attest to the important role parents play in providing access, encouragement, and modeling active lifestyles (Beets et al., 2010). Children are more likely to be physically active when their parents are physically active, include the children in their activities, and provide encouragement and support (Davison et al., 2006). In this study, young people who had parents who volunteered as coaches were five times more likely to be participating in OPA; a similar finding to the AusPlay study (Australian Sports Commission, 2018), in which 75% of children who had at least one parent participating in OPA were OPA participants compared to only 56% of children who did not have a parent engaged in OPA.

The importance of sibling participation has previously been examined in a systematic review which found that siblings can facilitate engagement in OPA by acting as role models, offering encouragement and support, and enabling vicarious learning experiences (Blazo and Smith, 2018). In a study examining constraints to sports participation for people with disability, Darcy et al. (2016) found that a lack of friends or companions to participate with and not wanting to participate alone significantly hindered participation. In the current study, young people who had a sibling participate in OPA were three times as likely to be current OPA participants. The presence of a familiar sibling may encourage participation by providing emotional and practical support.

The other external factor that was significantly associated with participation in the present study was school type. Students enrolled in mainstream schooling were more likely to be OPA participants than students enrolled in special schools or special developmental schools. This finding may reflect the influence of more severe disability in students attending non-mainstream schools as young people with higher support needs were found to face greater constraints to OPA participation (Mâsse et al., 2012; Darcy et al., 2016). Similarly, in this study, higher support needs, measured by the ability to understand simple instructions, were significantly associated with participation. Young people who could understand simple instructions were five times more likely to be OPA participants. In a Canadian study of participation in young people aged 5–14 (N = 145,180) with neurodevelopmental disorders and disabilities and chronic medical conditions, severity of disability was the most important child characteristic to hinder participation (Mâsse et al., 2012). Although no significant difference in participation according to disability type was found in this study, attending non-mainstream schooling and not being able to understand simple instructions were significantly associated with non-participation, suggesting that these young people may have greater support needs which act as a barrier to participation. Future studies examining the impact of support needs on participation in OPA are warranted.

The only internal assets that differed between current OPA participants and non-participants were the parent-reported strength of love of PA/sport and meeting PA recommendations. It is unsurprising that youth who love PA and/or sport are more likely to be participants given the previous finding that enjoyment is a key driver of participation in young people. What remains to be answered is how to cultivate this love of PA in young people with disability. As previously discussed, the influence of family (parents and siblings) in modeling active lifestyles, facilitating access to OPA, and offering encouragement and praise is invaluable. Additionally, it is important to foster feelings of competence (self-efficacy) which has consistently been found to be a determinant of PA participation (Heah et al., 2007; Bauman et al., 2012). In this study, self-efficacy was measured using the fPRC items relating to increased skill, independence, and confidence in performing the activity. After including items relating to involvement (motivation, happiness, social connection, persistence) and items relating to preference (importance, meaning), self-efficacy was no longer a significant predictor of participation. Nevertheless, young people of parents who endorsed the barriers "my child has difficulty performing the activities" and "my child finds the activities too challenging" were significantly less likely to be participating in OPA, indicating that self-efficacy is an important contributor to participation. Perceptions of self-efficacy may be particularly significant to young people with disability as parents have noted the frustration and loss of confidence their children felt when they compared their skill level with other participants without disability (Shields and Synnot, 2016). The benefits of participating in adapted physical activities where skills can be developed in a safe and supportive environment were highlighted in a recent study

(Nyquist et al., 2019). Children reported feeling comfortable learning new skills with other children with disabilities because they did not feel singled out or different. They also appreciated having sufficient time to develop mastery and felt optimistic that these newly acquired skills could be transferable to a mainstream OPA setting (Nyquist et al., 2019). Similarly, Shields and Synnot (2016) reported the need for inclusive pathways where children can progress from segregated activities through to mainstream or competitive sport.

Cultivating a love of PA in young people with disability by providing more supportive and enjoyable activities is an important way to assist youth to meet the Australian government's recommendation of 60 min of moderate to vigorous PA per day. Only 35% of the participants in this study met these guidelines which is similar to rates for Australian children in general (30% of children aged 2–17 met the PA guidelines according to the Australian Institute of Health and Welfare, 2018); however, it has been suggested that there is an "amplified" concern of inactivity for young people with disability due to an increased risk of obesity, social isolation, and mental health concerns (Anderson and Heyne, 2010). Young people in this study who were not currently participating in OPA were significantly less likely to be meeting these PA recommendations; therefore, finding ways to increase OPA participation by addressing the facilitators and barriers identified in this study could assist in meeting the recommended daily PA. In a recent Australian study of PA levels during OPA, participants typically spent 40–50% of a sport's practice session (e.g., soccer or netball) in moderate to vigorous PA as measured using an accelerometer (Ridley et al., 2018). The median duration of an OPA session was 1 h (Australian Sports Commission, 2018); therefore, one session would contribute to almost half the daily PA recommendation. In this study, 24% of the participants currently engaged in OPA participated four or more times a week, 53% participated two to three times a week, and 23% participated once a week. Current participation in OPA at these levels would not be sufficient to meet daily PA guidelines, and only 46% of the OPA group were meeting PA guidelines. Participation in OPA four or more times a week had the greatest odds of meeting PA guidelines; however, the cost to families engaging in OPA this frequently may be prohibitive.

There are a number of limitations in this study. Firstly, the sample included only three young people (1%) whose main language spoken at home was not English. This is significantly less than the 21% of Australians who speak a language other than English at home (Australian Bureau of Statistics, 2016). Seven young people (3%) were of Aboriginal or Torres Strait Islander heritage. This figure is representative of the Australian population (in 2016, Aboriginal and Torres Strait Islander people comprised 3.3% of the population according to the Australian Institute of Health and Welfare, 2019); however, compared to non-Indigenous Australians, Indigenous Australians are 1.8 times as likely to have a disability (Australian Institute of Health and Welfare, 2019). Future studies would benefit from ensuring the greater inclusion of young people whose main language at home is not English and Indigenous Australians. A further limitation relating to the sampling strategy was the use of predominantly online recruitment and an online survey. Online social media was the most frequently reported method of participants hearing about the study; however, families who have regular access to online material may not be representative of all families who have a child with a disability.

Additionally, the online survey was not previously trialed in the disability population, although survey items were derived from established models of participation (e.g., the fPRC), from a review of the OPA literature, and by consultation with a multidisciplinary team of health professionals. A power calculation was also not conducted due to limited research from which to estimate likely effect sizes. Instead, the sample size was based on pragmatic considerations, namely, the amount of data that could be collected without a significant increase in resources. Furthermore, during analysis, the five-point Likert scale responses were recoded into binary variables. Although this method can diminish power, many of the key relationships were significant; hence, if the five-point scale had been maintained, the relationships would be more likely to be significant. Consequently, this limitation does not compromise our confidence in the key conclusions.

Moreover, while the sample included a diverse range of disabilities, 41% of parents reported the young person's primary disability to be ASD, consistent with data from the NDIS indicating that children on the autism spectrum currently comprise the largest primary disability category in Australia (National Disability Insurance Agency, 2018). Although there was no significant difference in OPA participation according to disability type, the over-representation of participants on the autism spectrum may have bearing on the types of facilitators and barriers that were endorsed as well as the internal and external assets reported. An additional limitation was the reliance on parent-reported facilitators and barriers. While we decided to collect information regarding barriers and facilitators to OPA engagement from the parent perspective to avoid over-burdening youth with the comprehensive list of barriers and facilitators we wished to investigate, other research involving young people with disability has successfully engaged young people in identifying facilitators and barriers (Heah et al., 2007; Shields and Synnot, 2016). Therefore, future studies comparing OPA participants and non-participants might benefit from including child-reported factors.

CONCLUSION

This study confirmed prior literature in reporting that young people with disability do not participate in OPA at the same rate as their peers without disability. This is concerning given the weight of evidence which supports the potential for OPA to improve physical and mental health and to foster positive youth development (Murphy and Carbone, 2008; Holt et al., 2017). What this study adds to the literature is the identification

of several factors that differentiate OPA participants from non-participants. Interventions to promote participation in OPA for young people with disabilities should firstly focus on ways to increase intrinsic motivation during OPA. Secondly, the experience of enjoyment is crucial for ongoing participation in OPA (Martin, 2006; Heah et al., 2007; Nyquist et al., 2016); therefore, interventions should focus on making OPA enjoyable. Thirdly, young people benefit when their family are also engaged in OPA. Interventions that promote participation of siblings and parents will facilitate participation of young people with disability. Finally, some young people are being hindered from participating due to a lack of financial resources. Supportive government policies to cover costs associated with OPA would lessen the financial burden on lower income families.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available due to ethical considerations.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Deakin University Human Research Ethics Committee 2016: 336. Written informed consent to participate in this study was provided online by the participants, and where necessary, the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

NP and CE were involved in recruitment and data collection. MW analyzed the data. All authors participated in the conception and design of the study, were involved in data interpretation and manuscript drafting, and read and approved the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2020.568723/full#supplementary-material

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The Generalized Adaptation Account of Autism

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The heterogeneous phenomenology of autism together with diverse patterns of comorbidities led in the past to formulation of manifold theories and hypotheses on different explanatory levels. We scrutinize most recent findings from genetics, neurobiology and physiology and derive testable hypotheses about possible physiological links between domains. With focus on altered sensory perception and neuronal processing in ASD, we assume two intertwined regulatory feedback circuits under the umbrella of genetics and environmental factors. Both regulatory circuits are highly variable between individuals in line with the heterogeneous spectrum of ASD. The circuits set off from altered pathways and connectivity in ASD, fueling HPA-axis activity and distress. In the first circuit altered tryptophan metabolism leads to higher neurotoxic substances and reinforces the excitation:inhibition imbalance in the brain. The second circuit focuses on the impact and interaction with the environment and its rhythms in ASD. With lower melatonin levels, as the pacemaker molecule of the circadian system, we assume misalignment to outer and inner states corroborated from the known comorbidities in ASD. Alterations of the microbiome composition in ASD are supposed to act as a regulatory linking factor for both circuits. Overall, we assume that altered internal balance on cellular and neurophysiological levels is one of the main reasons leading to a lower ability in ASD to adapt to the environment and own internal changing states, leading to the conceptualization of autism as a condition of generalized imbalance in adaptation. This comprehensive framework opens up new perspectives on possible intervention and prevention strategies.

Keywords: autism, adaptation, multicausal pathogenesis, connectivity, circadian rhythm, tryptophan, environment, stress

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INTRODUCTION

The challenge of autism research to comprehensively unify the array of symptoms in social interaction and communication as well as repetitive and restricted interests and behaviors (American Psychiatric Association [APA], 2013) is unmet. Moreover, autism spectrum disorders (ASD) are characterized by extreme phenomenological heterogeneity. Genetic research in the past decades has shown large concordance rates (Folstein and Rutter, 1977), while the exact genetic mechanisms causing ASD remain elusive, with over 170 candidate genes associated with ASD known to date (SFARI Gene, 2019). Meanwhile, cognitive, neurobiological, endocrinological and environmental theories have been formulated, with each respective level furthering our understanding of ASD but not being

able to explain the etiology of symptoms on other levels. As a consequence, giving up on a single explanation of autism has been suggested (Happé et al., 2006). A multicausal pathogenesis converging to the spectrum of autistic phenomenology seems likely. Nevertheless, we believe that a theoretical framework of ASD attempting to unify most recent state-of-the-art findings from diverse levels of explanation can create a synergistic understanding of ASD in its whole complexity.

At this moment, several new leads are being followed in autism research that renew our thinking about neuronal connectivity in ASD (Tomasi and Volkow, 2019), gene x environment interactions (Rossignol and Frye, 2012) and involvement of the gut microbiome (Sarkar et al., 2018; Xu et al., 2019) opening many new questions, in particular on the links between discussed domains.

Thus, here we review the state-of-the-art knowledge in several current key domains of autism research, spanning genetic signaling pathways of neurodevelopment, neuronal connectivity and thalamic filter mechanisms, circadian rhythms, immunology, social functioning, neuroendocrinology, and the gut-brain-axis. We propose viable links between the key domains, generate targeted hypotheses and put forward a comprehensive framework of ASD that allows for the graded phenomenological expression observed across the spectrum.

MAIN ARTICLE

Alterations of Neurodevelopmental Signaling Pathways in ASD

Incontestably, ASD is highly heritable and based on a complex genetic etiology. In the latest GWAS the polygenic heterogeneity of autism-subtypes is confirmed qualitatively and quantitatively (Grove et al., 2019). *De novo* mutations, especially copy number variants (CNVs) and gene disrupting point mutations, which are supposed to have a larger effect in ASD, contribute to the individual liability, <5% (Gaugler et al., 2014; Iossifov et al., 2014), far less compared to the overall heritability. Special emphasis should be placed on the recent identification of five risk loci for ASD and seven additional loci that are shared with other traits (Grove et al., 2019).

With a view to converging pathways, genes of the WNT signaling pathway (Kalkman, 2012; Mulligan and Cheyette, 2016; Kumar et al., 2019) as well as calcium signaling and the MAPK signaling pathway are widely associated with ASD (Wen et al., 2016). KCNN2, as a voltage independent Calcium-activated potassium channel, represents a highly significant locus in the genetics of ASD (Grove et al., 2019). Activation of KCNN2 modulates neuronal excitability by membrane hyperpolarization, potentially boosting the risk of an altered excitation/inhibition ratio between neurons. Thus, these genetic alterations likely have a negative effect on intracellular and intercellular communication leading to altered connectivity via synaptic plasticity.

The WNT signaling pathway helps coordinating neurodevelopmental processes like cell proliferation, synaptogenesis, polarity and differentiation (MacDonald et al., 2009). WNT3 as one of the 19 ligands of the WNT signal cascade has been reported to be elevated in the prefrontal cortex of ASD patients (Chow et al., 2012). WNT2, as another ligand, is important for cortical dendrite growth and dendritic spine formation, while alterations of dendritic spines result in neurodevelopmental diseases (Oliva et al., 2013). Prostaglandin E2 as an inflammatory molecule is known to strengthen the canonical WNT-pathway (Wong et al., 2014).

The specific genetic architecture of ASD is still unknown. The interconnection of rare *de novo* mutations and inherited variants of different genes in aspects of transcription and protein networks in ASD, might result in abnormal concentrations of neuroligins, altered interconnection and synapse formation, dysregulation of the excitation/inhibition ratio as well as impairments of the immune system, referring to immune cell activation by Calcium as a core molecule. Moreover, we assume that the heterogeneous spectrum of ASD is amongst others caused by an underlying gradual effect of genetic alterations, while their dysregulation gets reinforced by a proinflammatory profile leading to a vicious circle.

As a major effect of these negative feedback mechanisms, we propose individuals with ASD to suffer from a reduced capacity to physiologically adapt to inner and outer states leading to a dysfunctional homeostasis. This imbalance is affecting the whole organism, as will be spelled out in detail for each building block in the following sections. ASD is thus proposed to be understood as a condition of generalized imbalance in adaptation.

Connectivity in ASD

Hypothesis 1: Local thalamic underconnectivity and long-range overconnectivity leads to chronic distress.

In several resting-state functional magnetic resonance imaging (rfMRI) studies in the last years, mostly all based on a relatively small sample size, heterogeneous results were found with respect to local and long-range connectivity in ASD that lead to the hypothesis that ASD might present with more local and less long-range connectivity compared to non-autistic people (Belmonte, 2004; Anderson et al., 2011). Results were equivocal though and several rfMRI studies demonstrated long-range overconnectivity between brain regions (Monk et al., 2009; Di Martino et al., 2014; Cerliani et al., 2015). In a recently published study a large number of rfMRI datasets of individuals with ASD (n = 565) were compared with datasets of unaffected healthy controls (HC; n = 605) using functional connectivity density mapping (Tomasi and Volkow, 2019). The anterior thalamus showed *local underconnectivity*, while *increased long-range connectivity* of the whole thalamus was observed with several cortical sensory areas (Tomasi and Volkow, 2019), correcting previously assumed characteristics of connectivity in ASD.

The anterior thalamus is a brain structure that contains the ventral anterior and the dorsomedial nuclei with their projection to the prefrontal cortex and to primary/association visual, auditory and somatosensory cortical areas (Behrens et al., 2003). With growing age this area showed an increase of local functional connectivity density (lFCD) in both groups, ASD and HC, but significantly less so in ASD (Tomasi and Volkow, 2019). The degree of local connectivity reduction in the anterior thalamus

compared to HC was positively associated with symptom severity in ASD (Tomasi and Volkow, 2019). Local connectivity correlates positively with the brain glucose metabolism, which reflects activity state and energy demand of the brain (Tomasi et al., 2013). The whole thalamus showed higher functional connectivity with the insula, somatosensory, motor, premotor and auditory areas and the middle cingulum for ASD compared to HC (Tomasi and Volkow, 2019). These neuroanatomical areas are associated with core symptoms of ASD: social impairment is linked to the *temporal sulcus*, language and communication dysfunction to the *thalamus/superior temporal sulcus/premotor cortex* and repetitive, stereotyped behavior to the *thalamus* and *motor areas* of the cortex amongst others (see Amaral et al., 2008).

Hence, the thalamus seems to be a key region for understanding ASD neuropathology given no other brain region with significant findings of connectivity abnormalities between HC and ASD patients was found in this large sample (Tomasi and Volkow, 2019). On the one hand the *thalamus* is mainly responsible for filtering information for regulated consciousness and alertness. It also integrates sensory and motor signals (Bell and Shine, 2016). Simplified, what passes through the thalamus comes to our awareness. The anterior thalamus with its observed local under-connectivity (Tomasi and Volkow, 2019), leads us to the assumption that in this region, that assesses sensory information of different qualities with respect to their importance of transmission, local communication and activity between neurons is impaired or disrupted. There is no clear evidence whether only the excitatory or inhibitory system or even both are affected in the anterior thalamus due to the macroscopic methods used (Tomasi and Volkow, 2019). It would be plausible though that both systems are affected in a quite individual way.

Many details of sensory information might pass through this physiological filter, with the whole thalamus showing increased projections to several brain areas. Increased long-range connectivity to different sensory areas might be an explanation for sensory abnormalities in ASD, such as hypersensitivity or sensory overload (O'Neill and Jones, 1997; Bromley et al., 2004; Harrison and Hare, 2004). In keeping with this line of thought is the report of abnormal resting states in EEG in ASD (Wang et al., 2013) that might be caused by the increase of long-range connectivity and could explain the *signaling imbalance theory* relating to elevated excitation and reduction of inhibition in brains of people with ASD, as well as the association of ASD with epilepsy (Croen et al., 2015), which is defined as a disorder of neuronal hyper-excitation. Furthermore, long range overconnectivity of the thalamus could account for autonomous nervous system (ANS) dysfunction in ASD (Panju et al., 2015). ANS dysfunction is proposed to be related to sympathetic hyper-arousal and a lower parasympathetic tone, shown by an increased heart rate, larger tonic pupil size and decreased heart rate variability (HRV) (Bal et al., 2010; Daluwatte et al., 2013; Porges et al., 2013; Kushki et al., 2014; Panju et al., 2015), what can be seen as symptoms of distress.

In fact, neuronal hyper-excitation and the associated chronic distress is in line with a whole series of findings of somatic complications found increased in ASD. For instance, increased neuronal activation in the CNS and ANS due to dysfunctional abnormalities of thalamocortical connectivity might explain why sleeping disorders are commonly found in ASD (Aldinger et al., 2015). Sleep is highly controlled by the circadian clock system where adaptation to the surrounding environment, like day and night, is fundamental. Associated with sleep disorders are gastrointestinal disturbances (GID), which are likewise commonly found in ASD (Klukowski et al., 2015). Children with ASD are often affected with autoimmune disorders, allergies, GI disorders, sleep disorders and seizures (Croen et al., 2015), while adults with ASD often suffer from chronic medical conditions, including dyslipidemia, hypertension, diabetes, obesity and thyroid disease (Croen et al., 2015). Especially the prevalence of stroke and Parkinson's disease, as well as vitamin deficiency is also significantly increased in individuals with ASD (Croen et al., 2015).

The Circadian Clock in ASD

Hypothesis 2: Abnormal neuronal connectivity via altered excitation (glutamate)/inhibition (GABA) leads to dysregulated melatonin synthesis affecting the circadian rhythm and genetic transcription.

With respect to the large prevalence of sleep disorders around 50–80% (Richdale and Schreck, 2009; Souders et al., 2009; Mazzone et al., 2018) in ASD, several studies investigated melatonin or melatonin metabolites showing abnormalities in ASD (Rossignol and Frye, 2011). Melatonin is an endogenous neurohormone transmitted mostly by the pineal gland, synthesized from serotonin in a two-step pathway. The common amino acid synthesized into serotonin and melatonin is tryptophan (see **Figure 1**).

Melatonin is important for the circadian clock in mammals. Beside its function in regulation and adjustment to exogenous stimulation, by day and night, it has an important role as antioxidant. Its immunomodulatory function is unclear, but there is the concept of melatonin as an "immune buffer" that has an anti-inflammatory compound during acute inflammation (Carrillo-Vico et al., 2013). On the assumption of the immunosuppressive role of melatonin and its decreased concentration in ASD, many individuals with ASD should show abnormalities in their immune system and its peripheral immune cell concentrations (Rossignol and Frye, 2012; Malkova and Hsiao, 2016), as well as gastrointestinal inflammatory diseases. Indeed, all of these are known comorbidities highly prevalent in ASD (Croen et al., 2015).

The key brain region associated with melatonin is the nucleus suprachiasmatic nucleus (SCN) that gets activated by different pathways of the visual system. Synthesis and release of melatonin in the pineal gland and retina follows a circadian rhythm. The SCN or melatonin itself is regulating many other circadian clock dependent systems, such as temperature, blood volume, behavior, locomotor activity, water balance, metabolic and immune functions (Bass and Takahashi, 2010; Mohawk et al., 2012; Scheiermann et al., 2013; Curtis et al., 2014; Labrecque and Cermakian, 2015). The aim of the body circadian clock is to synchronize rhythms and gene expression to a constantly changing environment in order to save homoeostasis in the whole

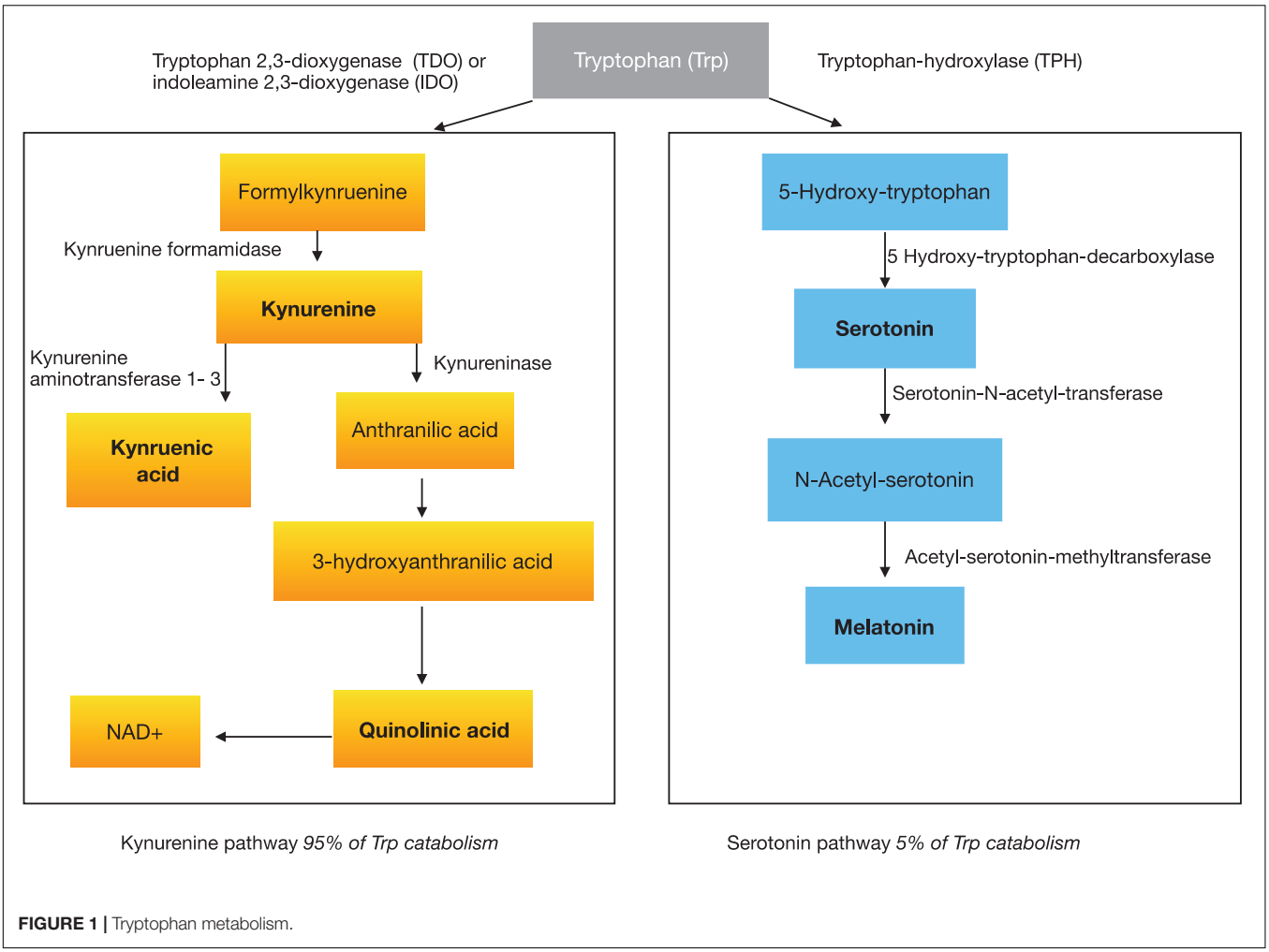

FIGURE 1 | Tryptophan metabolism.

organism. In order to achieve this kind of inner balance and efficient cellular responses, synthesis of melatonin and its binding to receptors, as well as receptor sensitivity, needs to be well regulated (Dubocovich et al., 2003).

Regulating factors are substances such as vasoactive intestinal peptide (VIP), neuropeptide Y, opioids, GABA, dopamine and glutamate (Dubocovich et al., 2003). If glutamate as an excitatory neurotransmitter and GABA as an inhibitory molecule are in an abnormal relation to each other, as mentioned in the *signaling imbalance theory* of autism, regulation of melatonin synthesis gets affected. Glutamate is an important modulatory molecule that inhibits melatonin synthesis by decreasing its genetic expression and the activity of its key enzyme (Villela et al., 2013). High glutamate levels might be one factor amongst others causing lower melatonin concentrations in ASD (Rossignol and Frye, 2011).

Melatonin is also meant for regulating synaptic plasticity (Frank, 2016). Periodic waves of the GABAergic inhibition in the hippocampal circuits are provided by the SCN (Frank, 2016). Dopaminergic synapses of the striatum show plasticity, while dopamine synthesis and metabolism is following a rhythmic expression due to the direct transcriptional link of dopamine gene activation by the core clock (McClung, 2007; Parekh et al., 2015).

Findings that melatonin production in adolescents and young adults with ASD is lower compared to HC (Tordjman et al., 2012) leads to the assumption that the circadian clock system is also affected in autism. Dysregulation of the circadian clock system and its clock genes might be caused (Nicholas et al., 2007, 2008) not mainly from genetic mutations. A lack of resting and sleep disturbs the normal shaping process of synaptic connections (Frank and Cantera, 2014). If the inner-circadian clock system gets disrupted, gene transcription and translation is necessarily affected negatively. For instance, abnormally low melatonin concentrations in ASD intensify sleep disorders and abnormal synaptic plasticity in the brain via dysregulation of neurotransmitters. This effect is bidirectional and can reinforce the impairment of the circadian clock system. Sleep quality affects the ANS and the immune system. Studies about the circadian clock function and its effects on our behavior describe the phenomenon of jet-lag, a misalignment of internal circadian rhythms and external time (Comperatore and Krueger, 1990; Waterhouse, 1999). Symptoms resulting from jet-lag are insomnia (Arendt, 2009), decreased alertness

and impaired cognitive skills. Chronic jet-lag is supposed to cause depressed mood, reduced psychomotor coordination and gastrointestinal disturbances (Waterhouse et al., 2007). Many of these symptoms also fit the comorbidities found in ASD. Chronic jet-lag in rodents was shown to lead to an increased risk of cardiomyopathies (Penev et al., 1998) and early death (Davidson et al., 2006), risks that are also found to be markedly increased in individuals with ASD (Croen et al., 2015).

The SCN synchronizes peripheral oscillators in several organs via hormonal and neuronal pathways. In one organ different subgroups of clock genes exist distinguishable on the basis of their transcription rate and velocity. Different periodic rhythms exist directly besides each other controlled by the main pacemaker, the SCN, and the hormone melatonin. Therefore, temporal disorganization of the circadian system during jet-lag likely disrupts overall physiological coordination. Indeed, reduction of melatonin-functioning was found to correlate with the severity of ASD symptoms (Rossignol and Frye, 2011).

The Immune System in ASD

Hypothesis 3: Chronic distress, sleep disturbance and disruptions of the circadian clock system and melatonin homeostasis result in increased cortisol concentration and immune system disarrangement. Due to the assumption of a bidirectional pathway, we understand this as a self-reinforcing process in ASD.

Disruption of the circadian clock system and sleep plays a critical role in immune system homeostasis (Castanon-Cervantes et al., 2010). Innate and adaptive immune responses are regulated in a time of day-dependent manner (Haspel et al., 2020) Melatonin, a potent antioxidant, is known to have pleiotropic effects on the immune system (Carrillo-Vico et al., 2013). Glutamate's inhibitory effect on melatonin synthesis involves interactions between astrocytes and pinealocytes, through the release of astrocytic TNF-alpha, a potent mediator of inflammation (Villela et al., 2013). TNF-alpha, a proinflammatory molecule, stimulates amongst others the release of corticotropin-releasing hormone (CRH) from the hypothalamus (Watanobe and Takebe, 1992). CRH activates via ACTH the secretion of glucocorticoids, like cortisol. High levels of neuronal glutamate might therefore not only decrease melatonin levels in the SCN, moreover it elevates inflammatory molecules via paracrine interaction with astrocytes and elevates immune system activity.

Typically, cortisol as an inflammatory corticosteroid hormone gets upregulated in stressful times to protect the body. Other inflammatory markers such as C-reactive protein (hs-CRP), cytochrome P450 (CYPp450) and 8-hydroxy-2′-deoxyguanosine (8-OH-dG) are blood plasma biomarkers related to inflammation and oxidative stress that were shown to be increased in ASD (Rossignol et al., 2014). Indeed, a higher prevalence of immune dysfunction is found in children with ASD (Croen et al., 2015). Moreover, the disruption of the circadian clock has an effect on the immune system as well because of its regulation of circadian clock genes in the adrenal gland where glucocorticoids, such as the hormone cortisol, are secreted.

Chronic distress is caused physiologically by the reaction of the HPA-axis and the ANS. Their hyperactivity can lead to several other disorders (Mc Ewen, 2006). For instance, a pathological HPA-axis functions as a predictor for cardiovascular diseases as well as for type 2 diabetes (Rosmond and Björntorp, 2000). Both are two somatic comorbidities significantly increased in autistic individuals (Croen et al., 2015). Of course, this dysregulation pathway is not characteristic or specific for ASD but it is quite important to mention, given melatonin being an antagonist of cortisol. In general, if melatonin is low in ASD patients, as shown above, cortisol gets upregulated. Several findings support the concept of abnormalities in stress response in ASD also at the cellular level (Essa et al., 2013; Rossignol et al., 2014). Reduced antioxidant defense is reported in several neurological diseases (Essa et al., 2013). There is high evidence that an increase of oxidative stress has also an impact on the pathology of ASD (Essa et al., 2013). Markers of oxidative stress correlate with ASD severity (Rossignol et al., 2014). Moreover, there is the assumption that observed oxidative stress is a chronic condition in autistic individuals (Rossignol et al., 2014). Several studies have reported an elevated production of oxidative markers, an increased exposure to environmental pro-oxidants and a decrease of antioxidant in ASD (Essa et al., 2013). Abnormally low antioxidant levels index a low functioning oxidative stress response. A significant increase of an oxidative stress marker, lipofuszin, is reported in three language areas of autistic people compared to controls (López-Hurtado and Prieto, 2008), while other studies were able to show higher immunoreactivity in several brain areas of ASD individuals (Rossignol et al., 2014). These findings lead to a higher secretion of free radicals with their potential to damage various structures of human brain and to influence CNS development negatively. Oxidative stress is not only interesting in times of brain development, rather it is a factor with impact on cell and membrane integrity, excitotoxicity and energy metabolism (Essa et al., 2013), dynamically in interaction with environmental factors and molecules of the immune system.

The immune system, as a link between genes and environment, is assumed to be affected in ASD (Ashwood and Wakefield, 2006; Rossignol and Frye, 2012). Therefore, due to the individual amount of severity of neurological, somatic and genetic abnormalities, flexible adaptation to the environment and an adequate stress response down to the cellular level, is hampered, with impact on a cognitive and psychological level. This is congruent with an understanding of autism as a condition of generalized imbalance in adaption.

Social Functioning and Oxytocin in ASD

Hypothesis 4: Higher levels of stress, with a low functioning oxidative stress response and elevated cortisol concentration in ASD cause downregulation of oxytocin secretion and gene expression and increased methylation of the OXTR gene, overall with impact on social behavior and interaction.

Social interaction is often reported as being stressful for autistic people (Corbett et al., 2010). The physiological correlate for stress is the activity of the HPA axis and its secretion of ACTH and cortisol.

Several studies have investigated cortisol levels and its circadian rhythm in autistic individuals. An elevation of fetal cortisol concentration has been reported (Baron-Cohen et al., 2015) with potential impact on early CNS development. Moreover, there is evidence that children with autism show a more variable cortisol rhythm and a significant elevation of cortisol following exposure to a novel, non-social stimulus (Corbett et al., 2006). Further investigations found a higher serum cortisol response, with significantly higher peak cortisol levels and prolonged duration and recovery of cortisol elevation following a stressor in ASD (Spratt et al., 2012). Cortisol levels during a peer-interaction task and after the game differed significantly between ASD and TD children, with higher levels in the ASD group (Corbett et al., 2016). Higher physiological arousal during playing was associated with heightened sensory sensitivity and increased stress in autistic children (Corbett et al., 2016). These findings lead to the assumption of an increased reactivity of the HPA axis to stress and novel stimuli in autism with a higher cortisol level measured peripherally.

Furthermore, cortisol is important in understanding the physiological role of oxytocin. Oxytocin is meant to modulate the stress response, by regulating cortisol and cytokine concentration inversely (McQuaid et al., 2016). Given that central oxytocin administration reduces stress-induced corticosterone release and anxiety behavior (Windle et al., 1997), leads to the conceptualization of an existing antagonism between the concentrations of oxytocin and cortisol in the CNS. Much research is done to study the question of correlation of autistic behavior and dysregulated oxytocin concentration (Modahl et al., 1998; Al-Ayadhi, 2005). Oxytocin is a neuropeptide produced in the hypothalamus and released by the posterior pituitary gland that plays a role in social bonding, childbirth and sexual reproduction. Social bonding is impaired, while the prevalence of anxiety is increased (Croen et al., 2015) in individuals with ASD suggesting less sensitivity to oxytocin caused by an abnormality in oxytocin receptor (OXTR) density during an early life period (Freeman et al., 2018). Accordingly, increased OXTR methylation in specific promoter regions (Gregory et al., 2009), as an effect of epigenetics, is in line with lower expression of the OXTR (Kusui et al., 2001) in ASD.

Thus, on the basis of a cascade of reduced local inhibition and increased long-range connectivity, with an assumed early-lifetime impact on the developing brain, leading to hyper-activity in the cerebral cortex and increased levels of cortisol, the proposed framework offers an account of lowered oxytocin as indeed observed in ASD (Modahl et al., 1998). This does not rule out an additional genetic coding dysfunction for oxytocin as well as for glucocorticoids (Brkanac et al., 2008; Patel et al., 2016).

Neuroendocrinology in ASD

Hypothesis 5: Higher inflammatory signaling molecules such as glucocorticoids in ASD shift the ratio from reaction pathways of tryptophan in favor of kynurenine rather than serotonin. This leads to another imbalance with the result of too much kynurenine, low tryptophan and serotonin as well as, referring to further reactions, low melatonin.

The synthesis pathway of melatonin leads us to the amino acid tryptophan, which is essentially converted to serotonin in the first step of melatonin synthesis (see **Figure 1**). If there would be a lack of serotonin or tryptophan in the brain in the first instance, then not enough substrate would be available for further reactions to melatonin in the pineal gland.

Tryptophan is an essential amino acid, which must be supplied in the diet (Le Floc'h et al., 2011), usually representing a component of protein. Once absorbed from the gut it can exist free or albumin-bound in circulation. Tryptophan can cross the blood-brain-barrier (BBB) and takes part in the synthesis of serotonin in the central nervous system (CNS). There is evidence that individuals with ASD have low tryptophan concentrations peripherally (Kałużna-Czaplińska et al., 2017). Serotonin itself cannot cross the BBB, even though more than 90 percent is located in enterochromaffin (EC) cells of the gastrointestinal tract (Gershon and Tack, 2007). A lack of central tryptophan would lead to less serotonin as well as lower melatonin concentration in the brain.

Tryptophan is converted in a first step to 5-hydroxytryptophan (5-HTP) by the rate-limiting enzyme, tryptophan hydroxylase (TPH) (see **Figure 1**). Two isoforms of this enzyme exist, TPH1 and TPH2. They are both in different kinds responsible for the serotonin-synthesis in the enteric nervous system (ENS) and CNS. In the second step 5-HTP is converted to serotonin. Tryptophan gets dominantly transformed by the kynurenine pathway. Kynurenine is produced from tryptophan by two different enzymes: tryptophan 2,3-dioxygenase (TDO) and indoleamine 2,3-dioxygenase (IDO) (see **Figure 1**). TDO can be induced by glucocorticoids or indeed tryptophan itself. IDO is affected by certain inflammatory stimuli, such as IFN-gamma.

Hypothesis 6: We propose that a lower concentration of kynurenic acid and a higher concentration of QUIN via the increase of oxidative stress and the release of glutamate aggravates the imbalance of the ratio of excitation/inhibition in the brain and has a neurotoxic effect.

Kynurenine itself is metabolized along two distinct pathways. The first one leads to the production of the neuroprotective kynurenic acid (a7 nicotinic acetylcholine receptor antagonist and N-methyl-d-aspartate (NMDA) receptor antagonist at glycine site) while the second arm leads to the neurotoxic quinolinic acid (NMDA receptor agonist) (see **Figure 1**).

Peripheral measurements showed an imbalance in homeostasis of the kynurenine pathway products with higher levels of QUIN in autistic children (Gevi et al., 2016) and lower levels of kynurenic acid (Bryn et al., 2017), while the ratio between kynurenine and kynurenic acid was significantly higher in the ASD group (Bryn et al., 2017). This ratio reflects a neurotoxic potential. Abnormally high concentrations of QUIN in the CNS of individuals with ASD might be also caused by higher levels of its substrate kynurenine. QUIN is a neurotoxic molecule. It increases oxidative stress by elevating the production of free radicals as well as increasing glutamate release and inhibiting its reuptake by astrocytes (Tavares et al., 2002). The

latter aspect results in an elevated concentration of glutamate, leading to overstimulation of NMDA receptors, that cause disturbances in intracellular Ca2 + -signaling by weakening the sarco/endoplasmic reticulum Ca2 + ATPase (Fernandes et al., 2008). Elevation of glutamate by QUIN might have the potential to aggravate the excitation:inhibition imbalance in brain.

Consequences on intracellular signal cascades are also in line with alterations of genetics, like calcium and MAPK signaling pathways (Wen et al., 2016). These several influencing factors might intensify abnormalities in intracellular communication by long-term adaptation to this altered intracellular state. Moreover, we assume that altered internal balance on cellular and neurophysiological levels is one of the main reasons leading to a lower ability in ASD to adapt to the environment and own internal changing states.

The Gut-Brain Axis in ASD

Hypothesis 7: Alterations of microbiome composition in ASD weaken the availability of tryptophan peripherally and cause disturbances in the endocrine balance by maladaptation of feedback loops and generally misbalanced adaptation to the environment.

Brain and gut communicate through the gut-brain axis, where serotonin is meant to be a linking molecule (Fattorusso et al., 2019). The gut microbiota, a complex of bacterial community located in the GI tract, has been found to be essential for maintaining metabolic and immune health (Lynch and Pedersen, 2016). There is even more evidence that the composition of the microbiome influences brain development, neurogenesis and interacts with the ENS and CNS via the gut-brain axis. Bacteria have been found to have the capability to produce a range of major neurotransmitters, also known under the term "*microbial endocrinology*." Gut microbes are known to regulate the serotonin concentration in the blood and colon (Yano et al., 2015) via their production of short-chain fatty acids (SCFAs). SCFAs can also modulate the activity of the host's sympathetic nervous system (Kimura et al., 2011). By using a variety of preclinical strategies, it has been established that manipulating the composition of the gut microbiota across the lifespan or altering the trajectory of microbial colonization of the gastrointestinal tract quite early in lifetime influences the availability of tryptophan (O'Mahony et al., 2015). Interestingly, animal studies have shown that early life time distress leads, beside the observed dysbiosis in the microbiome, also to an increase of immune system and HPA axis activity (O'Mahony et al., 2009). These alterations are meant to persist over lifetime and have adverse effects on behavior, such as on regulation of the stress neurocircuitry, emotions and cognition.

Changes in the composition of the microbiome, called "*microbial dysbiosis*," have been reported in ASD (Van Sadelhoff et al., 2019). We assume that this alteration of composition is linked to an individually reduced availability of tryptophan in general, resulting in low tryptophan levels in ASD (Kałużna-Czaplińska et al., 2017). The reduced availability of tryptophan increases the activation of the sympathetic nervous system by SCFAs, consistent with our assumption of increased ANS and HPA activity. And consequently, the serotonin synthesis is upregulated peripherally in EC cells via bacterial metabolites, congruent to observed elevated serotonin concentrations peripherally in ASD, called hyperserotonemia (Hranilovic et al., 2007). Thus, we need to assume weakened feedback loops of tryptophan metabolism in ASD on the basis of an altered microbiome composition.

Hypothesis 8: Impaired genetic signaling pathways reduce intestinal epithelium barrier integrity aggravating proinflammatory state in ASD.

Moreover, cellular signaling cascades, like the WNT pathway, do exist not just in the CNS. WNT signaling is also important as a regulator in the intestinal mucosa by organizing epithelial stem cell identity and maintenance (Moparthi and Koch, 2019). Mutations of genes of the canonical WNT pathway might therefore result in lower intestinal epithelium integrity (Pinto et al., 2003). As a consequence, pathogens of the daily environment and metabolism have the opportunity to enter cells more easily and harm the host more effectively, leading to an increase of proinflammatory molecules by upregulation of the host's immune system and HPA-axis activity for defense.

Hypothesis 9: Alterations of microbiome composition in ASD aggravate the dysregulation of the tryptophan metabolism by increasing kynurenine levels peripherally and centrally, in line with weakened feedback loops in ASD.

Central effects of serotonin are related to the circadian rhythm, motor control, body temperature, vascular tone and cerebellar regulation, while in the gastrointestinal system serotonin regulates pancreatic, intestinal and gastric secretion, gastrointestinal motility and colonic tone. While tryptophan can cross the BBB, its availability is necessary for the amount of serotonin in the brain. Several gut bacteria can modulate the metabolism of tryptophan into kynurenine. Depending on the bacteria involved, kynurenine biosynthesis can be increased or decreased. Some probiotics have been shown to reduce kynurenine levels (Desbonnet et al., 2008), for instance. Given the microbial dysbiosis in ASD we assume that this alteration in the gut increases the concentration of kynurenine peripherally via the tryptophan metabolism and in the CNS via the BBB. Hence, the peripheral shift of the tryptophan metabolism toward more kynurenine and reduced serotonin is mirrored in the CNS (see Hypothesis 5).

Furthermore, through an increase of the proinflammatory state in the gut (see Hypothesis 8) the enzymes of the kynurenine pathway get upregulated, another self-reinforcing process in the periphery.

Finally, elevated kynurenine levels lead us back to the neurotoxic effect of QUIN and the decreased neuroprotective potential of kynurenic acid (see section "Neuroendocrinology in ASD"), further aggravating the previously explicated effect of an activity increase of ANS/HPA via altered connectivity in ASD (see section "Connectivity in ASD"), on the basis of genetic (see section "Alterations of Neurodevelopmental Signaling Pathways

in ASD") and environmental modulators (see sections "The Circadian Clock in ASD" and "The Gut-Brain Axis in ASD").

The Model – The Generalized Adaptation Framework of Autism

The proposed framework of ASD as a condition of generalized imbalance in adaptation can be subdivided into two intertwined negative feedback circuits (see **Figures 2**, **3**, respectively) under the umbrella of genetic alterations and the environment. Both feedback circuits are highly variable between individuals in line with the quite heterogeneous spectrum of ASD.

Following the first circuit (see **Figure 2**), WNT, calcium and MAPK signaling pathways are negatively affected in ASD and epigenetically relate to gene × environment interactions. Abnormal signaling cascades lead to alterations in the formation of synapses, intracellular communication and the excitation/inhibition ratio as well as to increased levels of neuroligins. On this basis we think that the findings of long-range overconnectivity and local underconnectivity of the thalamus in ASD lead to a lower filter function of information in the brain. The resulting simultaneous activity of different cortical areas causes higher activity of the ANS and HPA-axis and consequently increased secretion of proinflammatory molecules and glucocorticoids. These molecules intensify the kynurenine pathway of the metabolism of tryptophan (see **Figure 2**). Kynurenine thereby gets upregulated in its concentration leading to higher concentrations of QUIN in the brain. Kynurenic acid is lower and in relation to kynurenine it has a neurotoxic effect. QUIN increases the release of glutamate in the CNS in line with an imbalance of excitatory and inhibitory neurons. The neurotoxicity might also result in graded levels of cognitive impairments. Importantly, it also results in a reinforcing process by activation of the immune system for defense. Higher concentrations of proinflammatory stimuli could raise the level of kynurenine as a positive modulator of the two enzymes tryptophan-dioxygenase and indoleamine-2,3-dioxygenase. We assume that mismatched feedback loops are existent in this circuit.

The second circuit (see **Figure 3**) again starts off from altered signaling pathways and connectivity in ASD with higher HPA-axis activity and stress levels (see **Figure 3**). As mentioned, we propose that the kynurenine pathway is upregulated by response to higher stress levels in ASD. This results in a shift of the balance of the two possible reaction pathways of tryptophan and lower serotonin and melatonin concentration in the CNS. Lower melatonin leads to disturbances in the circadian clock system causing sleep disorders and higher

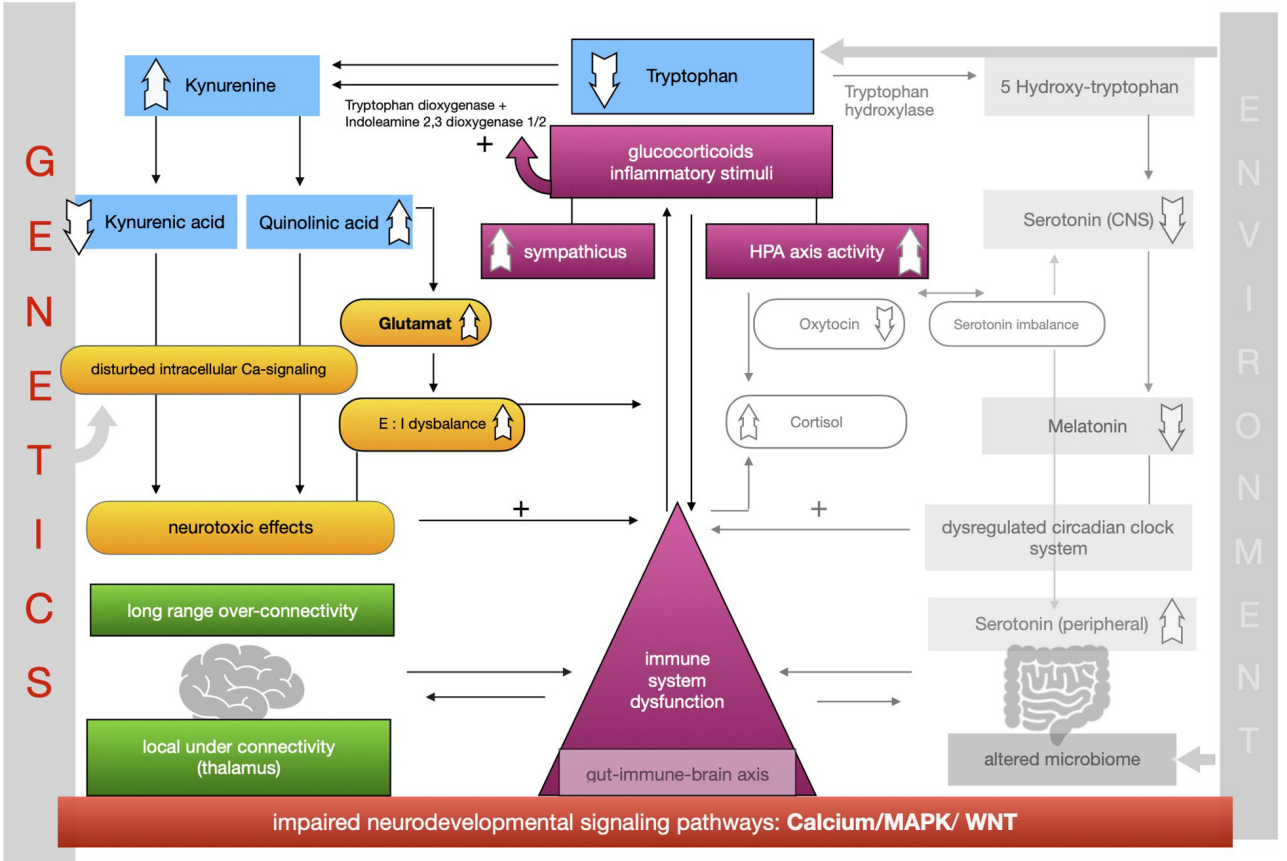

FIGURE 2 | Feedback circuit A.

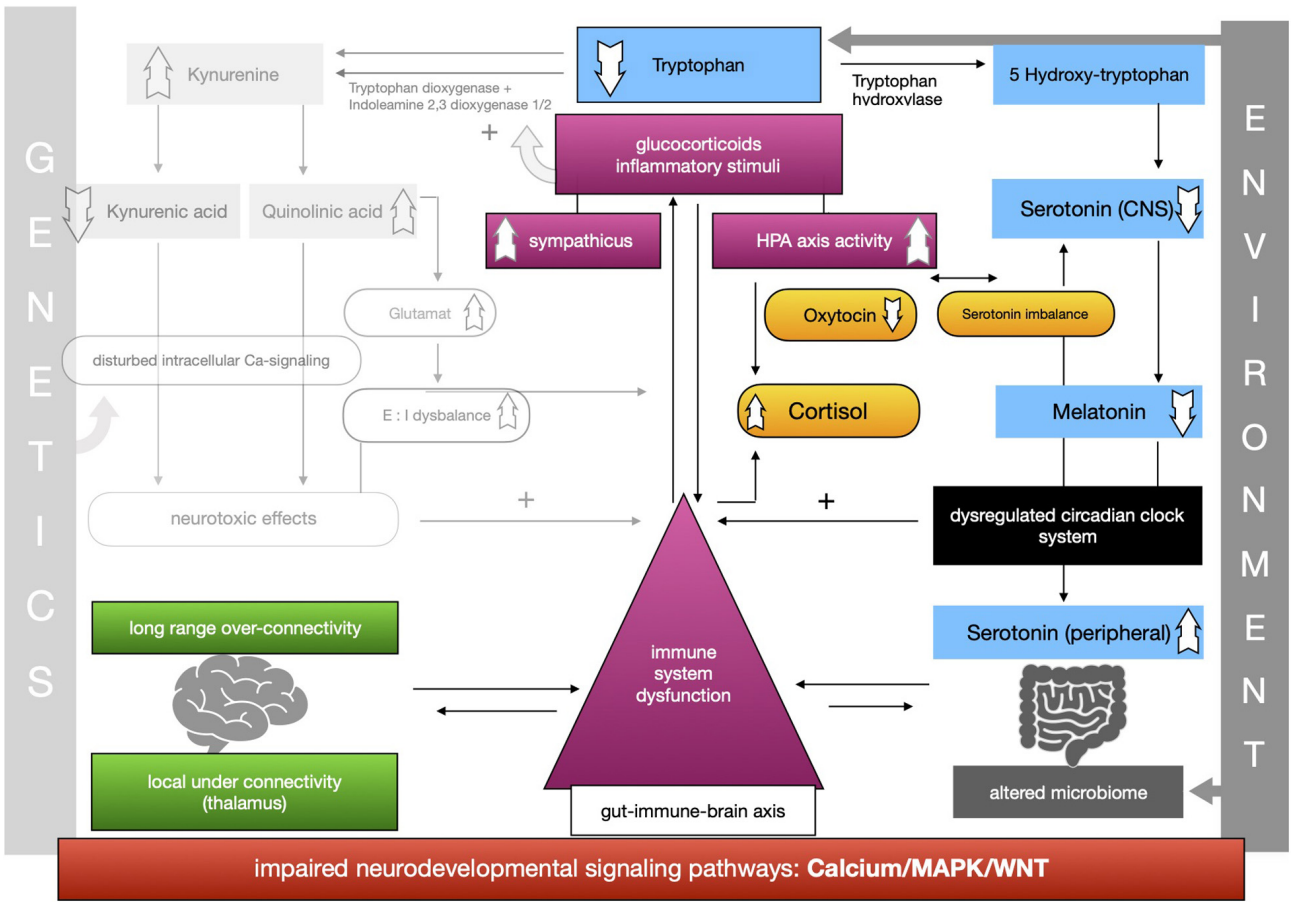

FIGURE 3 | Feedback circuit B.

distress and enhancing the activity of the immune system. The imbalance of excitatory/inhibitory neurons, as presented in the first circuit, aggravates the dysregulation in melatonin synthesis via their regulatory effects. The circadian clock system stands as an example for the impaired adaptation ability to the environment in ASD. The relationship between melatonin and cortisol, a typical stress hormone, is antagonistic. Here too there is a reinforcing process. The HPA-axis hormone oxytocin is antagonistically reduced, which is in line with reported impairments of social bonding and interaction in ASD. Meanwhile, the environment impacts the intestinal system where alterations of the composition of the microbiome are commonly observed in ASD. Due to altered signaling pathways, we assume that intestinal and BBB integrity is weakened so that more pathogens and other toxic substances can enter cells more easily and reinforce the activity of the immune system. In this second circuit (**Figure 3**) we likewise propose weakened feedback loops leading to maladaptation of inner cellular and hormone pathways, especially in the gut-brain axis and its linking molecule serotonin. Microbial alterations lead to lower levels of tryptophan peripherally, while serotonin concentration is increased due to the stimulation of EC cells by SCFAs, a product of bacterial metabolism. Moreover the microbial dysbiosis in ASD might strengthen the imbalance of tryptophan metabolism in the CNS and gut by increasing the levels of neurotoxic kynurenine, leading to decreased serotonin as well as tryptophan concentrations in the brain. Higher activity of the immune system caused by the decreased epithelium barrier integrity reinforces the kynurenine pathway by enzyme stimulation.

Both circuits are intertwined into one whole self-reinforcing process in ASD, which is the basis of a generalized impairment of adaptation to the environment and one's own internal states. A comprehensible way to adapt to the lack of homeostasis is stereotypical and repetitive behavior, as an early learned regulatory self-stimulation that helps people with ASD in situations that are experienced as stressful due to the fact that this kind of behavior requires less adaptability.

Symptoms and comorbidities linked to ASD can be implemented within the theory of impaired adaptation (see **Figure 4**).

The Specificity of Maladaptation in ASD

While all neuropsychiatric conditions might have elements of maladaptation to the environment (e.g., sleep disorders are widely associated with several neuropsychiatric conditions), there

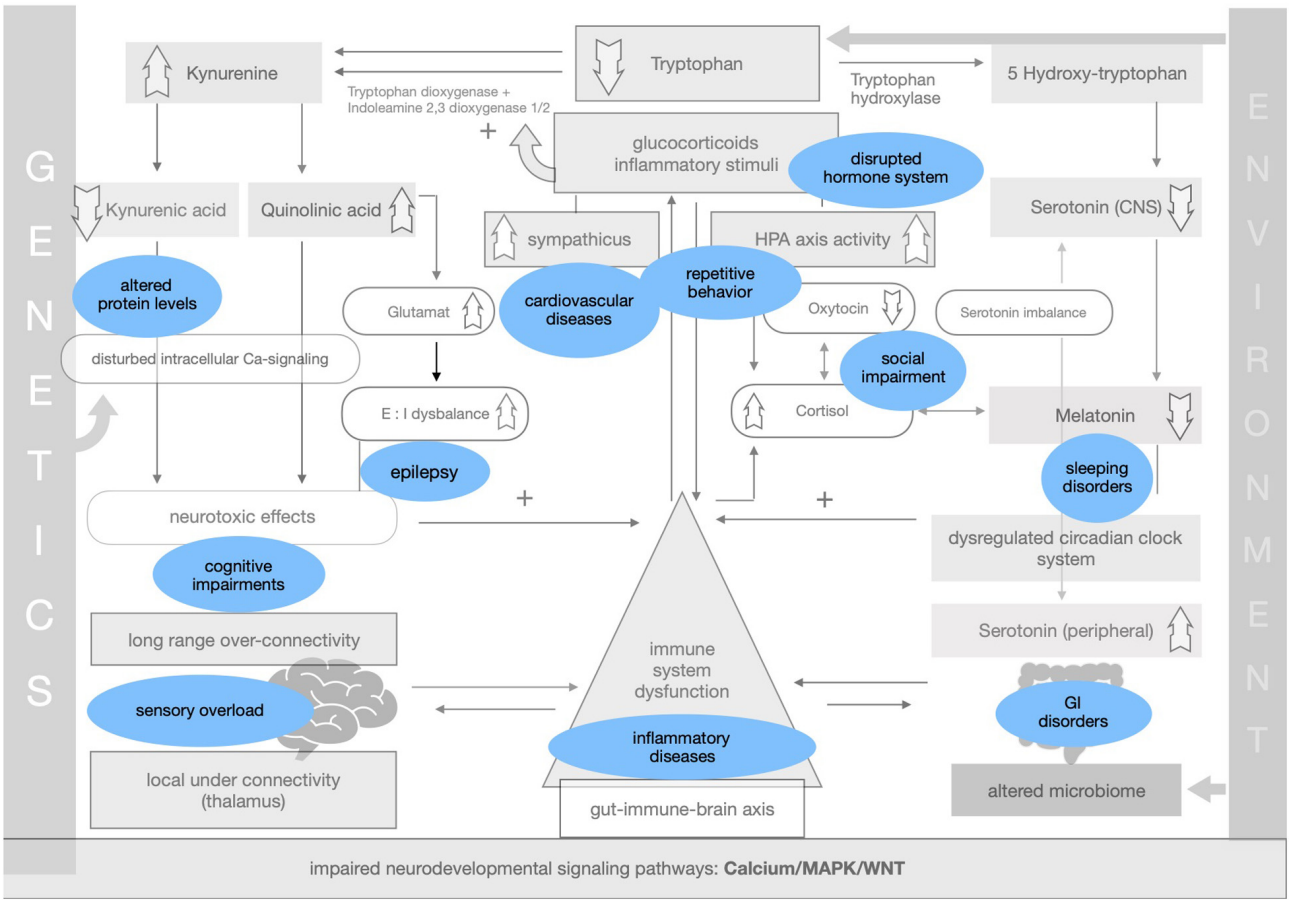

FIGURE 4 | Symptoms of ASD in the generalized adaptation framework.

BOX 1 | Outlook on potential treatment options in the generalized adaptation framework.

One suggested treatment plan would be to focus more on nutrition, especially in the subgroup of autistic children, to avoid obesity and associated medical comorbidities like cardiovascular and metabolic diseases. Constant blood screening of autistic people for inflammatory biomarkers might be helpful to see whether they are elevated or not and how long inflammation takes. Even more there should be also the focus on screening levels of corticosteroids, like cortisol and its metabolites, due to their suggested antagonistic effect with melatonin and oxytocin.

Drug treatment in general should carefully consider co-occurring effects on the circadian clock and the sympathetic nervous system. Based on the thesis of neuronal hyperactivity the aim should be to reduce the level of activation of the sympathetic nervous system relatively in ratio to the parasympathetic one in order to care for good sleep, lower stress level, lower concentrations of inflammatory markers as well as for lower risk suffering from epileptic attacks and cardiovascular disorders. Relieving the HPA-axis and the ANS via the sympathetic pathway might be an effective treatment in future times for ASD symptoms.

A possible drug for sleeping disorders could be melatonin, especially in childhood. Several studies could show quite good evidence for improved sleep parameters and better daytime behavior (Rossignol and Frye, 2011). The focus lays on the attempt to resynchronize the circadian clock to environmental stimuli so that gene transcription and translation work more regularly and that regulation of all several subgroups of clock genes in the different tissues are not disturbed that much as without melatonin treatment because of the primary pacemaker function of melatonin in the chronobiology system.

Propranolol, a non-selective beta-blocker, is inhibiting the noradrenaline and adrenaline system. After oral administration, it gets absorbed up to 90% in the liver. Usually it is used for treatment in hypertension and angina as well as migraine. Contraindications are bronchial asthma and bronchospasm because of increasing these symptoms. Propranolol is lipophilic and enters the BBB, so that it gets used for treating anxiety disorders. Propranolol reduces autonomic dysregulation by blocking the sympathetic nervous system. Therefore, it can be useful for treating disorders concerning to emotional and behavioral deficits caused by hyperarousal. Sixteen reports are found in a review about the use of propranolol in ASD (Sagar-Ouriaghli et al., 2018). The results from the eight single-dose clinical trials led to significant improvements in cognitive performance, improvement in semantic networks and functional connectivity. The remaining eight single case reports and case series showed improvements in anxiety, aggressive, self-injurious and hypersexual behaviors. In no study a negative observation has been reported so far for the treatment with this kind of beta-blocker apart from high dose treatment that caused hypotension (Sagar-Ouriaghli et al., 2018). This can be seen as a treatable side effect. It should be mentioned that autistic individuals with high dysregulation in the autonomous nervous systems and low functional connectivity gained the greatest benefit from propranolol treatment.

Further research should be done beside the use in clinical practice of propranolol, whether there are other lipophilic beta-blocker molecules that have a similar effect and are suitable for ASD treatment.

are two important characteristics differentiating maladaptation in ASD from that potentially to be found in other neuropsychiatric conditions. First, deviant adaptation to the environment in ASD is arguably present from birth, given the neurodevelopmental nature of ASD, in contrast to late acquisition of potential deviant adaptation in neuropsychiatric conditions, such as depression. Maladaptation during early neurologically vulnerable phases of development would arguably strongly shape individual developmental pathways. According to the neuroconstructivist perspective (Karmiloff-Smith, 2006) we need to take the ontogenetic development into account that is continuously forming the microconnectivity of the brain and the fine-tuning of functional circuits. Importantly, the neurodevelopmental perspective with cascading effects of constricted adaptation throughout levels of functioning *per se* entails the generalized nature of deviant adaptation.

Second, deviant adaptation to the environment in ASD would differ from that potentially found in other neuro*developmental* disorders, in that in the latter case it would be confined to specific aspects of brain development and neurological functioning. For instance, although adaptation problems are clearly observable in Attention-Deficit/Hyperactivity Disorder (ADHD), these are primarily confined to executive functioning. In contrast, in ASD deviant adaptation is thought of as a pervasive process in that it is thought to affect states of metabolism, neuronal connectivity, cognition, immune system, social interaction, and individual somatic levels. This neurodevelopmental pattern of pervasive deviant adaptation combined with the incapacity of a physiological transformation process during development is a specific pattern within ASD.

Thus, the presented framework points toward the importance of environmental factors to be adapted to each individuals' needs and symptom severity to reduce negative somatic effects. In addition, compensatory strategies have to be learnt, and this learning should be supported by tailored interventions, to cope with challenging situations and thereby improve health and life expectancy of autistic people in general.

CONCLUSION

The proposed framework seeks to unify most recent findings on neurobiological, endocrinological, cellular and connectivity levels in order to explain association with and gradation of various symptoms of ASD and its comorbidities. The presented model accounts for the phenomenological heterogeneity of the spectrum. The feedback circuits provide the opportunity to alleviate stress reactions, the activity of the immune system and consequently the risk of comorbidities by taking care of dynamical changing environmental factors in each individual case. The theory has the potential to give an explanation why there are also autistic individuals with mild symptoms and a lower risk for comorbidities in line with higher lifetime quality. Concerning repetitive behavior as a possible compensatory strategy in ASD to deal with these several imbalances, the model also highlights strengths of autistic people. The multivariable conceptualization of ASD in the proposed framework as a generalized adaptation imbalance declares, why no one specific key treatment for autistic symptoms can be established. While deviant adaptation is not specific to ASD, the pattern of pervasive deviant adaptation on all the levels described in the framework is argued to be specifically characteristic for ASD - a theoretical framework which should be subject to targeted future research (see **Box 1** as an example for future research topics).

AUTHOR CONTRIBUTIONS

CG conceived the theory. All authors critically discussed the results and contributed to the final manuscript.

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Interests and Strengths in Autism, Useful but Misunderstood: A Pragmatic Case-Study

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Background: Studies on autistic strengths are often focused on what they reveal about autistic intelligence and, in some cases, exceptional and atypical reasoning abilities. An emerging research trend has demonstrated how interests and strengths often evident in autism can be harnessed in interventions to promote the well-being, adaptive, academic and professional success of autistic people. However, abilities in certain domains may be accompanied by major limitations in others, as well as psychiatric and behavioral issues, which may challenge their inclusion in support programs.

Objectives: To provide an in-depth, pragmatic, real-life example of the psychological and psychiatric management of interests and strengths in an autistic adolescent.

Method: An autistic teenager, C.A., with above-average calendar calculation and musical abilities, received psychiatric, neuropsychological, and language standardized and clinical assessments, combined with a measurement of his musical and calendar calculation abilities. C.A. and his parents then received psychiatric and psychological support over a 14-month period, targeting their perceptions of C.A.'s interests, strengths, and co-occurring difficulties.

Results: C.A. had a verbal IQ within the intellectual disability range and a non-verbal IQ in the low mean range. Modest calendar calculation, absolute pitch, and matrix abilities coexisted with severe receptive and expressive language disorder. The discrepancy between his abilities in areas of strengths and his limitations in other domains led to anxiety, frustration, and sometimes behavioral issues. Displacing the focus from academic performance to interests, as well as promoting the use of his strengths to develop new skills independently of their short-term adaptive benefits yielded positive effects on C.A.'s self-assessment, quality of life, and behavior at follow up.

Discussion: The appealing idea that abilities mostly found in autistic people, such as calendar calculation, can be directly harnessed into academic achievement and lead to paid employment may have detrimental effects, especially when such abilities are modest and associated with other limitations. These abilities should be primarily used to maximize well-being and quality of life, independently of their short-term adaptive function, which may or may not be positive.

Keywords: autism, case-study, adolescent, strengths, interests, calendar calculation, absolute pitch, intervention

BACKGROUND

Intense interests and special abilities in autism have been the subject of constant attention for almost a century (Feinstein, 2011). Originally only a subject of curiosity, they were quickly used as a gateway to autistic cognition, with the seminal studies of Hermelin and O'connor (i.e., 1971; 1975), Shah and Frith (1983), and Mottron and Belleville (1993, 1995). They were generally considered to have no adaptive or social utility and to be more useful to the scientists who study them than to the people who possess them. These abilities were often considered *islets of abilities* seen in so-called *idiot savants* (Feinstein, 2011), therefore not related to general intelligence, and even to this day they remain pathologized. For example, repetitive behaviors and specific interests are often considered to hinder learning in early intervention (e.g., Rogers and Dawson, 2009).

The notion of the exceptionality of abilities in autism and their uselessness as an adaptation tool both started to change at the beginning of the twenty-first century. First, a kinship between the outstanding abilities that one could find in savant autistic and non-savant autistic people was proposed (Heaton, 2009; Howlin et al., 2009; Meilleur et al., 2015). This supported the possibility that most autistic people would present an ability stronger than predicted by their apparent general level. Since then, the capacities and interests of autistics have been slowly reintroduced as valuable, at least to increase the quality of life of the people who possess them (Winter-Messiers, 2007; Chiodo et al., 2017; Davey, 2020).

Aside from this positive, but limited, trend in the scientific literature, certain aspects of this beneficial change of orientation regarding autistic capacities, associated with better acceptance of autistics as members of the human community, could have harmful side-effects for autistic people. First, employment based on skills can lead to an increase in the burden of what autistic people are exposed to in a professional environment, and therefore their anxiety to perform satisfactorily in the environment in which they work (Holmes and Annabi, 2020). Savant autistic abilities have sometimes been interpreted as suggesting that every autistic person has exceptional, innate capacities, allowing them to effortlessly learn and retain large amounts of data. This latter notion is now part of the popular image of autism, attested by television series and media stories. This unnuanced portrayal is seen as annoying by some parents (Happé, 2018). Strengths or talents, both in autism and within the neurotypical population, are to be distinguished from a gift as the latter implies that no effort or practice is needed to attain a certain level, which is not necessarily the case for a strength or a talent. Furthermore, strengths could have an adverse as well as a beneficial effect depending on context (Eigsti and Fein, 2013; Russell et al., 2019). Finally, the superiority of many autistic people in non-verbal tasks (Muth et al., 2014) and how they are underestimated by intelligence assessments (Courchesne et al., 2015, 2019) may hide that a normal distribution of intelligence is expected in autistic and non-autistic people. Uneven intelligence profiles, and general intelligence may obviously influence their learning abilities, both inside and outside their domain of expertise.

This case-study of an autistic teenager and his family focuses on the challenges arising from a direct association between the level of these strengths and expected academic performance or adaptive level. It does not question that autistic people have strengths, nor the fact that these strengths represent an advantage for themselves and the entire community.

CASE HISTORY

C.A. is the only child of a Mediterranean family who immigrated to Canada before their son's birth. C.A.'s cousin has Tourette syndrome, but no other neurodevelopmental conditions were reported in his relatives. His mother reported having had some learning difficulties in math and sciences. She could speak English and French but her level of comprehension was limited in both, whilst C.A.'s father had a better level in both languages. French is the language used at C.A.'s school, but he was exposed to his parents' native language, before he was exposed to French at the daycare center he attended from approximately 20 months to 4 years of age. From 4 to 5 years of age, he attended a specialized preschool in English. Motor development was delayed (sitting: 7 months, remaining upright: 12 months, walking: 20 months). After a first word at 12 months, he used only a few isolated words over a 3-year period.

C.A. was first assessed in a development clinic at 28 months of age following speech and language development concerns. Delayed onset of eye contact, apparent disinterest in other children, and hand-leading were noted. At that time, fine motor abilities were in the 12th percentile. Communication precursors were limited and speech assessment impossible due to lack of production. His adaptation to daycare routines and social demands was difficult. He also displayed an interest in vacuum cleaners, toys that made music, and cause-and-effect toys. The first combinations of two words were observed at

4 years of age. He, however, displayed early learning of the alphabet in French at around 2 years of age (for example, he compared the Mercedes logo to the letter M) and was categorizing objects by their shape at this age. The parents reported that he self-learned these skills through a cause-and-effect toy (e.g., the toy would say *Taxi* and C.A. would press the letter T). Audiology was within normal limits. Minor dysmorphic features (hypotelorism, soft and pliable ears, unilateral right clinodactyly, right single transverse palmar crease) were noted but considered to be non-clinically significant. C.A.'s first diagnosis was global developmental delay.

A second assessment took place when C.A. was 3 years 11 months old and included an ADOS (Lord et al., 2000). At that time, he produced two- to three-word non-grammatical sentences, together with stereotyped language. Toilet training had just been achieved. Eye contact was still atypical. As he was not testable by conventional tests, such as the Weschler scales, he was administered the *Eye-Hand coordination*, *Non-verbal reasoning*, and *Language* subscales of the Griffiths Mental Development Scales, normed from birth to 8 years of age. C.A's performance on all subscales showed a significant developmental delay, of 18 months on average. For language, the delay was superior to 24 months. He received a diagnosis of autism following this assessment. No strength-oriented tests were administered at that time and hence no more precise and domain-specific conclusions can be drawn regarding his intellectual level during childhood.

As for services and interventions received, when he was 2 years old, C.A. went to a family daycare twice a week. At three he attended private sessions focused on global motricity development. At four and continuing through childhood, he received specialized services in the community and in a specialized elementary school, in addition to private services in speech and language therapy, occupational therapy, and psychology. A neuropsychological assessment conducted around age 10 yielded a heterogeneous intellectual profile (which was also found in the current study, see below) and concluded to a visuo-constructional apraxia, attention and executive functioning difficulties. Between 11 and 13 years of age, he was followed in a specialized youth mental health clinic, and re-assessed. A co-occurring mild intellectual disability and ADHD diagnosis were given. The intellectual assessment conducted at that time yielded homogeneous results (between the first and 2nd percentile, except on fluid reasoning, which was at the 8th percentile). Adaptive behaviors were in the extremely low range, which led to the diagnosis of mild intellectual disability. He was prescribed atomoxetine (10 mg) and methylphenidate (40 mg) for ADHD symptoms, with an initial positive effect on attention. He was still taking this medication during the course of this study, which began when C.A. was 13 years 8 months old. However, C.A. and his parents reported not seeing overt effect of this medication anymore. At the end of the follow-up, his family doctor prescribed a different brand of methylphenidate and a dosage of 45 mg, without additional benefit.

C.A. currently attends a specialized class for autistic youths, without other support. His parents realized just before the beginning of this study that C.A. could calculate calendar dates. Moreover, they reported that he is good at recognizing songs and has absolute pitch according to his music teacher. They contacted the research team and decided to participate in the study seeking ways to potentialize C.A.'s strengths.

METHODS

The study was initiated when C.A.'s parents sought help from the research team for optimizing their son's calendar calculation and musical abilities. Research-oriented investigations were conducted conjointly with a multidisciplinary clinical team. Consent to use the results from the assessments and intervention for research purposes was obtained from C.A. and his parents at the beginning of the study and the Frontiers consent form for the publication of case studies was signed by the mother prior to manuscript submission. An initial psychiatric assessment session was conducted by a psychiatrist (L.M.), a psychologist (V.C.), and a psycho-educator (V.L.) with C.A. and his parents in the hospital setting at the very beginning of the study. This initial meeting was followed by an extensive cognitive, language, academic, adaptive, and psychosocial assessment, including an investigation of interests and strengths. The language assessment was conducted based on best practices for language assessment in autism (Broome et al., 2017) by a speech and language therapist (A.S-D.) while the other domains were assessed by V.C., V.L., and L.B. The assessment was conducted between 13 years 8 months and 13 years 11 months of age. **Tables 1**, **2** present the tests and subtests used for the cognitive, language, adaptive, and psychosocial assessment.

Three psychiatric, six individual psychotherapy, and six parental coaching sessions, as well as a mid-intervention summary session, followed the assessment sessions. These intervention sessions were conducted in order to answer the clinical needs identified during the assessment. Hence, their focus and duration were driven by clinical considerations (see below for more details about the content of the sessions). Both the father and mother were present for the initial assessment, the mid-intervention session and one of the parental coaching session. Only the mother was present for the other parental coaching sessions and the follow-up assessment. Clinicians kept detailed progress notes of each session, which were used to describe the interventions conducted. These notes were also used to document barriers, setbacks and progress observed by the clinician at each session. A final follow-up session, during which some questionnaires were re-administered, was conducted 4 months after the end of the intervention. These are preceded by an «*» in the following descriptions and measure the primary (Quality of Life) and secondary (Adaptive Behaviors) outcomes targeted by the intervention.

Cognitive, Language, and Academic Assessment (13:8–13:11 Years Old)

Raven's Standard Progressive Matrices (RPM: Raven et al., 1998)

The RPM is a measure of fluid intelligence. Its administration does not require language; it is composed of five sets of 12 matrices of increasing difficulty. RPM have been shown to

TABLE 1 | Results for each test and subtest administered.

Measured function	Tests or subtests used	Percentile
IQ	RPM WISC-V	13 3 (FSIQ) 2 (Verbal comprehension) 3 (Visuospatial reasoning) 27 (Fluid reasoning) 8 (Working memory) 1 (Processing speed)
Expressive language	EOWPVT-4 CELF-CDN-F + WIAT-II	27 (Vocabulary) 0.2–4 (Oral expression)
Receptive language	EVIP-A CELF-CDN-F	<1 (Receptive vocabulary) 0.2 (Receptive language)
Reading	BALE+ *Le Vol du PC* WIAT-II+ *Le Vol du PC*	<0.1–2.3 (Decoding)[a] 0.1–3 (Reading comprehension)
Writing	WIAT-II+BALE WIAT-II	2.3–14 (Spelling)[b] 2 (Grammar and written expression)
Adaptive behaviors	VABS[c]	4 (Adaptive behavior composite) 4 (Communication) 7 (Daily living skills) 5 (Socialization)

[a]*Decoding of regular words was within the average. Reading speed for single words was within the average.* [b]*Spelling for regular words was within the average.* [c]*Completed by mother.*

be suited to the assessment of autistic intelligence, especially when verbal skills are limited (Courchesne et al., 2015). It was administered to C.A. in a single session.

Wechsler Intelligence Scales for Children—Fifth Edition (WISC-V: Wechsler, 2014)

The WISC-V is the most widely used intelligence test. It provides information on visuospatial reasoning, fluid reasoning, verbal comprehension, working memory, and processing speed. All mandatory and supplementary subtests were administered to C.A. in two separate sessions.

TABLE 2 | Questionnaires administered and results.

Questionnaire	Construct measured	Results
PedsQL™ 4.0	Youth's quality of life (parent rated)	62% satisfied
	Physical	93.75%
	Emotional	45%
	Relation with peers	50%
	Studies	60%
	Youth's quality of life (self-rated)	56.25% satisfied
	Physical	90%
	Emotional	35%
	Relation with peers	45%
	Studies	55%
FQoL	Family quality of life	
	Family interaction	4.33/5–satisfied
	Parenting	4.00/5–satisfied
	Emotional well-being	3.25/5–neutral
	Physical/Material well-being	4.20/5–satisfied
	Disability-related support	2.75/5–neutral
DASS-21	Mother's mental health	Invalid
Parenting style and dimensions questionnaire	Self-reported parenting style	Authoritative
Parenting sense of competence	Self-reported parenting efficacy and satisfaction	High
HIBOU	Sleep issues screening	5/27 (Normal)

Clinical Evaluation of Language Fundamentals—French Canadian Version (CELF-CDN-F: Wiig et al., 2009).

The CELF is a comprehensive test battery that assesses language abilities and was found to be representative of spontaneous speech in children with autism (Condouris et al., 2003). It was used here to provide information on both receptive and expressive language. It was administered to C.A. during one of the assessment sessions conducted by the speech and language therapist.

Wechsler Individual Achievement Test—Second Edition (WIAT-II: Wechsler, 2005)

The WIAT-II assesses academic achievements of children and adolescents. It provides information about their level in reading, written language, oral language, and mathematics. For the purpose of the present study, all subtests included in the reading and writting subscale were administered, as well as the oral expression subtest. It was also administered as part of the language evaluation and provided information regarding C.A.'s academic level in language-related subjects.

Expressive One-Word Picture Vocabulary Test—Fourth Edition (EOWPVT-IV: Brownell, 2000)

The EOWPVT-IV is a widely used expressive vocabulary assessment in which the participant is asked to name the pictures he is presented. This test was also part of the language assessment.

Échelle de Vocabulaire en Images Peabody (French Version of the Peabody Picture Vocabulary Test) (EVIP: Dunn et al., 1993)

The EVIP is the French version of the Peabody Picture Vocabulary Test, a receptive vocabulary test, in which the participant has to choose the one picture among four that best illustrates the word said by the experimenter. It was administered to C.A. by the speech and language therapist during language assessment.

Le Vol du PC (Boutard et al., 2006)

Le Vol du PC is a short story designed to assess reading speed, errors, and comprehension in youths aged from 11 to 18 years

old. This was also part of the language assessment and served to assess academic level in French reading.

Batterie Analytique du Langage Écrit (BALE: Jacquier-Roux et al., 2010)

The BALE is a test battery assessing written language level in children from second to fifth year of elementary school (7–10 years old in a typical curriculum in Quebec). The text-reading speed and accuracy, as well as the regular/irregular and non-word reading subtests, were administered to asses reading ability. The regular/irregular and non-word spelling subtest was used to assess spelling ability. As the BALE norms are relative to the academic curriculum of each grade and C.A. was pursuing fourth grade level French in school, despite not being age appropriate, the BALE corresponded to his current level in school. It was administered to C.A. as part of the speech and language assessment.

*Vineland Adaptive Behavior Scales—Second Edition (VABS: Sparrow et al., 2005)

The VABS is a measure of adaptive functioning. It provides information on functioning in the following areas: communication, socialization, daily living skills, and motor skills. This test was administered to the mother during the initial assessment phase and at follow up.

PSYCHOSOCIAL ASSESSMENT

*Pediatric Quality of Life Inventory Generic Core Scales (PedsQL: Varni et al., 2001)

The PedsQL^{TM} 4.0 is a 23-item scale that measures health-related quality of life in a multidimensional manner (physical, emotional, social, and school functioning) among children and adolescents (ages 2–18) using a 5 points Likert scale going from «Never a Problem» to «Almost Always a Problem». A mean level of satisfaction for each scale is derived from averaging the scores in this domain. It is used to document outcomes in clinical trials, including with autistic youths (Sheldrick et al., 2012; Safa and Islam, 2017). The French version of the Child Self-report and the Parent Proxy-report (ages 13–18) were used in the present study for initial and follow-up assessment.

*Beach Center Family Quality of Life Scale (Park et al., 2003), French Adaptation Directed by Chaume et al. (2019)

The Family Quality of Life Scale is a 25-item questionnaire that assesses satisfaction in five domains: family interaction, parenting, emotional well-being, physical/material well-being, and disability-related support using a 5 points Likert scale from very unsatisfied to very satisfied. It has been used with families of children with special needs (Boelsma et al., 2018) and autism (Hsiao et al., 2017). This test was also re-administered at follow-up.

Depression, Anxiety, and Stress Scale—21 (DASS-21: Lovibond and Lovibond, 1996)

The DASS-21 is a 21-item self-reported questionnaire to measure the severity of symptoms associated with depression, anxiety, and stress in adults, which is appropriate to evaluate these symptoms in parents of autistic children (Firth and Dryer, 2013; Lai et al., 2015).

Parenting Style and Dimensions Questionnaire (Robinson et al., 1995)

The Parenting Style and Dimension Questionnaire is a self-reported questionnaire that assesses parenting practices and categorizes them into authoritative, authoritarian, and permissive style.

Parenting Sense of Competency Scale (Johnston and Mash, 1989), French Adaptation by Terrisse and Trudelle (1988)

The Parenting Sense of Competency Scale is a 17-item questionnaire to assess parents' feelings about their parenting competency on a 1–6 Likert scale. Results vary from very low satisfaction to very high satisfaction. The questionnaire has been used with mothers of autistic youth (Tobing and Glenwick, 2007; Rodger et al., 2008).

OWL-Sleep-Inventory (HIBOU: Jaworski et al., 2016)

The HIBOU is a parent-reported questionnaire to screen for sleep problems in children. It is the French Adaptation of the BEARS (Owens and Dalzell, 2005).

INTERESTS AND STRENGTHS

Interests and Strengths Questionnaire for Preschoolers (ISQP)

The ISQP questionnaire (Larose et al., Submitted) was developed by experts in autism (including L.M., a co-author of this paper) and validated with autistic and typically developing children. It documents the strengths and interests of preschool-aged autistic children and their parents. It also includes questions on parental perception of the child's strengths and interests, and documents interventions that included or targeted the child's interests and/or strengths. It is composed of 19 multiple-choice and open-ended questions. The questionnaire was adapted and used as a parent semi-structured interview in the present study.

Absolute Pitch Assessment

Absolute pitch was assessed through the identification of 60 musical notes (Vangenot, 2000) separated into 6 musical dictations of 10 notes. Each note lasted 1,000 ms, followed by an ISI of 2,000 ms. No feedback was provided and there was more

than one octave between each consecutive note to prevent the use of relative pitch.

Calendar Calculation Assessment

We asked C.A. to identify the weekday of 10 past dates (from year 2000 to 2018) and 10 future dates (from year 2018 to 2037), one every 2 years. For each group of dates (past and future), the questions did not involve the same month more than twice and correct answers did not fall on the same weekday more than twice.

INTERVENTION (14:2–15:4 YEARS OLD)

Psychiatric Intervention

Three psychiatric intervention sessions were conducted by a psychiatrist (P.G.) in the hospital setting with C.A. and his parents. All sessions lasted between 1 and 2 h. The first two sessions were conducted conjointly with a nurse, who acted as the contact person for the family for the psychiatric intervention. The last session was a co-intervention session with the school pedagogy specialist, with the objective of better understanding the pedagogical objectives and alternative possibilities for school programs, based on C.A.'s interests.

Parental Coaching

Six parental coaching sessions were conducted with C.A.'s mother by a psycho-educator (V.L.). Three were conducted in the hospital setting and three at home. The father was also present in one of the session conducted at home. They all lasted between an hour and an hour and a half.

Psychotherapy With C.A.

Six individual psychotherapy sessions (three in the hospital setting, three at home) were conducted with C.A. by a psychologist (V.C). After the first three sessions of psychotherapy and parental coaching, a mid-intervention summary session took place with the parents, C.A., clinicians (V.C. and V.L.), and a psychiatrist (L.M.) to discuss the evolution of the situation and the intervention plan for the next sessions. The last three sessions were conducted in the home setting and ended with a part conducted conjointly with the parents, C.A., and clinicians.

Milieu Adaptation

Following the team's recommendation and discussion with the pedagogy specialist at C.A.'s school, a meeting was held between C.A., his parents, and the school pedagogy specialist at C.A.'s school to assess his interests and needs and discuss adaptations that could be implemented.

FOLLOW-UP (15:8 YEARS OLD)

A follow-up session took place 4 months after the end of the intervention. Based on the assumption that the follow-up discussion would be richer if conducted by clinicians familiar to C.A. and his parents, this session was conducted by V.L. and V.C., who respectively, conducted the parental coaching and individual psychotherapy sessions. This session included the re-administration of questionnaires assessing primary (Quality of Life) and secondary (adaptive behaviors) outcomes (see assessment section for details). An informal discussion about the family's experience throughout the study further documented the barriers and facilitators they encountered during the intervention. The discussion was conducted in part by V.L. and V.C with C.A.'s mother alone and in part by V.C. with C.A. alone.

RESULTS

Cognitive, Language, and Academic Assessment (13:8–13:11 Years Old)

C.A. presented overall intellectual functioning in the borderline range. However, his fluid intelligence assessed using the RPM was in the low average range, whereas his score on the matrix reasoning subtest of the WISC-V was in the 75th percentile, which is within the high average range for his chronological age, thus representing his better capacities. Furthermore, this high score on the Matrix subtests was drastically different from his score on the other subtest included in the Fluid Reasoning Index of the WISC-V: the Figure Weights subtest, on which C.A. obtained a score in the 5th percentile or borderline range. His score on the arithmetic subtest, assessing mental calculation abilities, was also in the borderline range around the second percentile. C.A. also presented dysprosody, depending on the topic discussed. Pronoun reversal was occasionally present. His expressive and receptive language level was significantly below that expected for his age group. His written language skills were consistent with what was observed orally. Despite relative strengths in specific domains of language, such as an expressive vocabulary within normal limits (low average range), C.A.'s language difficulties had a significant impact on his functioning and were consistent with the language ability profile of autistic youth with a co-occurring language disorder. His adaptive behavior level was in the borderline range. A higher score in motor skills is often observed in the VABS, as there is a ceiling effect for youths without a motor disorder. See **Table 1** for detailed results on cognitive and language assessment.

Psychosocial Assessment

Results from the quality of life questionnaires rated by the mother and the youth himself (see **Table 2** for details) indicated good physical health (around 90% satisfaction), which is similar to normative populations, whereas the emotional, social, and school domains were lower than his physical health satisfaction (varying from 35 to 60% satisfaction) and lower than what was reported in general population studies using this questionnaire (between 78 and 84% satisfaction) (Varni et al., 2003). His mother reported being satisfied or very satisfied with almost all aspects of their family life. She reported that her husband and herself were, however, not satisfied with the support and services received for their son at school and in the community, as her family is in need of someone to help them optimize their son's potential concerning his musical and calendar

abilities. His mother rated anxiety items on the DASS-21 as «doesn't apply to me, never», which led to the test being invalidated. Throughout the assessment the mother was reluctant to acknowledge any difficulties of challenges she was facing or to show vulnerability. Total parenting efficacy was within the mean and parental satisfaction was high. Behavioral problems were assessed using an adapted version of the tantrum questionnaire (Beauchamp-Châtel et al., 2019) to systematically document the frequency, intensity, and triggers of tantrums or meltdown. The results indicated that meltdowns occurred 1–3 times a week, for approximately 1–5 min each time, during the previous year. These meltdowns were mostly triggered when C.A. faced academic difficulties, causing him to hit his head with his palms, voice negative thoughts about himself ("loser," "stupid," etc.) and sometimes slap his mother (without hurting her).

Interests and Strengths

Interests: Youth's Report

C.A. reported interests in Lego®, videogames and YouTube videos. He mentioned that he liked listening to music but did not spontaneously mention playing music as one of his interests. He also mentioned two specific funny videos that he likes watching repeatedly on YouTube and watching documentaries about rappers.

Interests: Parent's Report

History of interests included moderate or elevated interests for dinosaurs, insects, animated characters, numbers, logos, trains, dates, toys with sounds, and electronic devices during childhood. More recently, the interests reported by his parents paralleled those reported by C.A., but also included his strengths, which were not identified by C.A. as interests. Indeed, the parents reported a high or intense interest in Lego®, but also in calendars, music, and books (biographies), which C.A. had not mentioned. When questioned about the amount of time spent on his interests, parents reported that he plays music four times a day for 5 min each, in the context of a course for which he has to do so. He never plays music for more than 15 min on his own. He prefers to play piano by rote memory rather than by reading musical scores. In contrast, he can spend up to 60 min flipping through books and searching Google, Wikipedia, or YouTube for information on the subject he is exploring. He can also spend more than 60 min playing videogames online. He spontaneously prepared trips by gathering information on the country to be visited. The parents reported being proud of their son's interests and press him to pursue them by encouraging him to read biographies or play music, for example, which again seem to be more related to strengths than interests.

Strengths: Youth's Report

C.A. reported having strengths in calendar calculation, geography, and music recognition, and being proud of these strengths.

Strengths: Parental Report

C.A.'s parents reported *relative* strengths (i.e., better than his overall general level of abilities) in reproducing constructions based on a model (Lego®), musical memory (identifying movies with the first notes of the soundtrack), spatial orientation, electronic-device manipulation, and calculations, and an *absolute* strength (i.e., better than what most people can do) in date memory/calculation. They reported no particular strength in reading, drawing, or puzzles. The parents reported being mostly positive about their son's strengths. They considered these as helpful to learning and not detrimental to daily activities. They also reported promoting their son's strengths when they identify one.

Strengths: Clinical and Empirical Assessment

Relative or absolute strengths associated with areas of interest were clinically explored. Concerning Lego® constructions, C.A. needed the help of an adult every few steps to correct errors and guide him and he was not particularly fast at completing the steps. Dates motivated him to expose himself to levels of language superior to his actual reading abilities. For example, he is interested in biographies and reported being focused on the dates. His interest in geography and politics led him to listen to the news on TV and search for information on the countries and cities he was going to visit. His apparent knowledge or understanding of politics were limited by his verbal level. For calendar calculation, C.A. was better for past dates (7 of 10 correct) than future dates (2 of 6 correct). The testing was interrupted because he expressed discomfort and anxiety when he could not provide an answer. The further away the dates were, the longer it took him to provide an answer. He reported basing his calculations on anchor dates. He remembered that movies are released on Fridays and computed his answers from the release dates of movies. He also mentioned that dates repeat every 6 years, which is not exact. His computational abilities for calendrical information was therefore in the modest range relative to other calendar calculators, but still above the average for the general population. For absolute pitch, C.A. was unable to complete the evaluation task in its original form and attempted to find the note's name by computing explicitly its distance from an anchor note. When notes were presented one by one, C.A correctly identified 5 of 10, which is still above chance for pitch recognition.

Psychiatric Assessment

C.A. was experiencing high levels of anxiety in his everyday life manifested by repetitive questions, sometime concurrent with behavioral issues. His anxiety comprised generalized anxiety themes (natural disasters, not having a seat in a plane) but was more often focused on pass/fail issues, such as academic success or the fear of being unable to become a financially independent adult. Although there was no indication of social anxiety, he pressured himself to succeed in school and in social interactions. Most of his repetitive questions on time schedules were related to school. Anxiety would rise quickly during any kind of assessment, when he did not know the answer, when the task difficulty increased, or when he realized he was not going as fast as his peers in an academic task. Unanswered questions, negative comments, or irritability when he did not understand something or faced an academic difficulty resulted in meltdowns.

He also reported self-depreciative thoughts, feeling discouraged about his language limitations and his learning difficulties, and voiced negative thoughts about himself ("I am a loser," etc.). He reported academic success as being of paramount importance to succeed in life and angrily attributed his academic challenges to his autism diagnosis, which for him encompasses all his challenges. His outbursts, which started when he was around 12 years old, circularly increased his anxiety and low self-esteem. He could calm down rapidly when his parents were able to reassure him and remind him of his strengths. He expressed feelings of shame and guilt for not being able to control them. Agreeing with him to alleviate this aspect of his anxiety resulted in more collaboration. He explicitly identified his best moments as non-school periods, holidays, and travel.

Intervention (14:2–15:4 Years Old)

The general goal of the intervention was to improve personal and family quality of life and improve C.A.'s general adaptation including, but not limited to, adaptive behaviors. The effects of the intervention were assessed at follow-up, 4 months after the end of intervention. The follow-up session therefore included administration of quality of life and adaptive behaviors questionnaires, but also focused on discussing the general well-being of C.A. and his parents.

Psychiatric Intervention

For C.A., the main axes of the intervention were the validation of his emotions and needs, re-explaining the school classification process, psychoeducation about the challenges associated with autistic signs vs. those associated. e.g., with a language disorder, and cognitive restructuring of the beliefs about how one can contribute to society and live a fulfilling life. For his parents, the intervention focused on promoting independence at home, encouraging them to limit their answers to C.A.'s repetitive questions (i.e., teaching them to reformulate their answers in several distinct terms, with the use of a visual support when possible). We also highlighted positively reinforcing good behavioral management, which they were able to sustain despite periods of increased school-induced stress or increased behavioral problems.

Parental Coaching

The focus of these sessions was to help C.A.'s parents in seeking learning opportunities that were suited for him, for example because they do not rely on language. They were encouraged to organize activities around his interests and strengths, regardless of their level or potential effect on their son's future. The sessions were oriented toward the acceptance of their son's limitations. The importance of pursuing pleasant activities, with no learning goal *per se*, was highlighted. Sessions also included stress and emotional regulation tools to help C.A.'s mother deal with her own anxiety and provide her with tools (emotional validation, use of the thermometer metaphor, breathing exercises) to better react to her son's anxiety and emotional outbursts. These sessions validated C.A.'s parents' efforts and devotion. Despite being reluctant to acknowledge the difficulties she was facing and the emotions accompanying these challenges, C.A.'s mother was deeply touched when validated in her parenting practices or in the emotions she could be facing.

Psychotherapy With C.A.

The goals of these sessions paralleled those of the parental coaching sessions. We provided C.A. with emotional regulation tools (i.e., deep breathing and visually tracking his stress level on a scale). Psychotherapy also focused on the understanding and acceptation of his limitations. We encouraged him to develop more autonomy so as to experience success in various domains and not just focus on academic achievement. He was also oriented to harness his strengths and pursue his interests through playful and pleasant activities with the psychologist (i.e., building with Lego, playing music, discussing movies, etc.).

Milieu Adaptation

For his current school year, C.A. was oriented toward a program to learn semiskilled trade jobs. One was to wash dishes and he reported hating it. He repeatedly said he wanted to pursue his academic education and learn new academic skills. He was first provided with self-taught didactic material to fulfill his interest in learning academic subjects. In parallel, the team coordinated with the school pedagogy specialist to re-orient him toward a program focused on academic subjects. At the end of the follow-up he had recently been moved to a different class and program in which more academic work was performed, but in which he still pursued work placement in manual jobs. This program change was deemed necessary so that C.A.'s school program would consider C.A.'s *interest* in school and academic subjects, regardless of his *level* in this domain.

Follow Up (15:8 Years Old)

All aspects of C.A.'s self-reported quality of life (physical, emotional, relationships, and studies), despite still being lower than population norms, showed increases at the follow-up assessment (see **Table 3**). During the follow-up discussion C.A. reported being happier in his new class, in which most of his friends from the previous year also were. He also reported liking the most recent job placement he had, disassembling electronics. He is now aware that completing a regular degree in college or university is not a realistic objective, but still has not figured out what type of occupation he would like to have after high school or if he would like to pursue his education in adapted programs. He was proud to succeed in doing some tasks that he previously thought he could not do by himself, such as cooking simple meals, and was working toward becoming more independent. He indeed wants to be able to live independently as an adult, but is not motivated to help around the house for now, as he is still young. He feels depressed about his communication difficulties and sees this as the main challenge in his acceptance of autism. He would like to have a girlfriend but is afraid his communication challenges will be a barrier to this aspiration. Overall, C.A. now has more realistic expectations concerning his strengths and he better understands his limitations. His self-assessment is therefore more accurate and he has an acute understanding of how he is regarded by others and how his own future could be challenging, which is painful for him. His mother perceives this as

TABLE 3 | Pre- and Post-intervention scores for primary and secondary outcomes.

Questionnaire	Construct measured	Pre–Post scores
FQoL	Family quality of life (/5)	3.70–4.60
	Family interaction	4.33–4.67/5
	Parenting	4.00–4.5/5
	Emotional well-being	3.25–4.75/5
	Physical/Material well-being	4.20–4.6/5
	Disability-related support	2.75–4.5/5
PedsQL™ 4.0 self-rated	Youth's quality of life (% satisfaction)	56.25–66.5%
	Physical	90–91%
	Emotional	35–50%
	Relation with peers	45–60%
	Studies	55–65%
VABS	Adaptive behaviors (percentile)	4th–3th
	Communication	4th–2th
	Daily living skills	7th–4th
	Socialization	5th–6th

a loss of motivation and hope and deplores this change. However, the family quality of life reported by C.A.'s mother has shown improvements, as has the self-reported quality of life.

According to the parental reports, the repetitive questions about his school schedule were at a tolerable level and the change of class was truly helpful for both C.A. and his parents. During an informal discussion about the study, C.A.'s mother stated that she appreciated having had a space to talk. She realized how important it is to emphasize things other than school and to enhance pleasant activities in her son's life. She stated examples of how she is now trying to promote his autonomy by asking him to help around the house with various tasks, although unsuccessfully. On a more negative side, C.A.'s measured adaptive behaviors had not improved. C.A.'s mother remained convinced that her son's abilities have been "given" to him for some purpose, and was unsatisfied by the intervention in this regard.

DISCUSSION

Summary of Findings

We presented here the case report of an autistic adolescent with modest abilities in calendar calculation and musical memory, adaptive behavior in the mild to moderate disability level, uneven task-dependent non-verbal IQ, and verbal abilities in the severe disability range. Interests, cognitive, psychiatric, and adaptive measures are reported, as well as psychoeducational, psychological, and psychiatric interventions and their short-term consequences. Illustrated by this case-study, we will now discuss the relation between relative and absolute strengths and general intelligence, and the positive and negative effects of the expectations grounded on them.

Interests, Strengths, and General Level of Intelligence

C.A's uneven profile is characterized, as is the case for many autistic people, by an important discrepancy between fluid intelligence and verbal abilities. He also presents domain-specific performances discrepancies: some areas (dates) and operations (calendar calculation) are performed at a much higher level than others (arithmetic). Therefore, intelligence cannot be deduced from his verbal and adaptive abilities, and is task-dependent. The measurement and practical use of his fluid intelligence are bounded to specific operations and materials, at least at time of assessment. His measured fluid intelligence is in the low average range in one test, but a peak in matrix reasoning on another one indicates that it could be underestimated (see below for more details). How this profile can be modified by access to new materials and education in general remains an open question, but this profile can be considered as characteristic of autism. Given that the level attained in the domain of expertise tends to increase with age and intelligence, strengths (both relative and absolute), despite being intrinsic to the autism diagnosis, may necessitate practice. The level attained can be experience- and intelligence-dependent.

Strengths in autism can be observed in individuals with superior intelligence, but also in individuals with lower intellectual potential. Peaks of abilities and general intelligence are not properly reflected by the concept of "islets of abilities" disconnecting them from general intelligence, and there is indeed a link between the level attained in the ability and *g factor* (general intelligence) (Hermelin and O'connor, 1986; O'Connor and Hermelin, 1988). C.A.'s abilities are among those that may reach an exceptional level within an autistic presentation. Although they exceed what most non-autistic and autistic people can do, they are of a modest level relative to other published savant abilities in the same domains (Mottron et al., 1999, 2006; Thioux et al., 2006; Bouvet et al., 2019). Therefore, the expectations for translating these strengths into direct and immediate adaptive outcome need to consider the context of domain/task-specificity but also the correctly measured general level of intelligence of the person.

The recent tendency to use interests and strengths to promote learning in autism should therefore be enriched by distinguishing between intense interests (being good at vs. being interested in), strengths (absolute, relative), intelligence and transferability to other domains. Recommendations to use interests were initially limited to using them as external reinforcements (e.g., Charlop-Christy and Haymes, 1998). More recent and rare recommendations suggest using the area of interest as learning material (Baker, 2000; Winter-Messiers et al., 2007; Courchesne et al., 2016; Ostrolenk et al., 2017). As for strengths in autism, they have long been included in interventions, with principles such as using visual support (Mesibov and Shea, 2010). This case-report suggests that the use of interests in intervention should be individualized as a function of the person's relative and absolute strengths, and transferability of abilities from the strong domain to other domains is not necessarily straightforward or doable at all. Following Dawson et al. (2008), these interests should at

least lead to making relevant material available to the person, and critically observing what happens next.

The Positive and Negative Effects of Expectations Based on Strengths

C.A. has anxious and depressive manifestations centered on academic success and his future as an independent adult, which leads to mild behavioral problems. His parents also reported preoccupations regarding their son's future, hoping that their son can find a way to use his abilities to learn academic skills. The pressure C.A. puts on himself to achieve better academic performance, in addition to the familial and institutional pressure to see immediate effects of his strengths on other areas of learning, contribute to generating anxiety. This results in intense frustration and behavioral problems when facing difficulties, damaging to his self-esteem and family life. It also contributes to the task-dependence of his cognitive performance, as this could explain the discrepancy found between the two versions of the matrix tests that he was administered (13th vs. 75th percentile). In the RPM (13th percentile), the difficulty, and therefore failures, increase within each of the five sets, in addition to increasing between the sets, and all items are administered regardless of the number of errors. In the Wechsler version (75th percentile), the increase in difficulty is constant and the test is stopped after three consecutive errors, which minimizes the total number of errors. Furthermore, throughout testing, a decrease in anxiety was observed when the administrator intervened, for instance by repeating that it was normal not to know all the answers. Hence, the context and C.A.'s state of mind may directly influence his ability to perform.

Interventions focused on helping him with his anxiety, expectations, and emotional regulation were conducted over a 14-month period with his parents, his school, and C.A. himself. These interventions attempted to shift the focus from academic success to academic learning, both at school and within the family. This focus did not fully correspond to the family's expectations and led to some frustration that the mother expressed to the team. Despite the fact that both C.A. and his parents were in search of a silver lining for their son's calendar calculation ability, to which the intervention did not answer, an improvement in C.A.'s well-being and behavior was seen at the 4-month follow-up and at the end of the intervention. However, we cannot be certain that the measured improvements will last, or that they were due to the intervention.

Adaptive Outcome vs. Quality of Life Associated With Interests and Strengths

C.A. is interested in learning and accumulating information about themes such as geography, movies, biographies, and dates, and has above average calendar calculation and absolute pitch abilities. Our observations indicate a potential risk of assuming a direct link between interests and strengths, and academic potential. This shortcut contributes to increasing the expectations for better academic performance, in turn increasing his anxiety and feelings of being a failure. Although C.A.'s interests and strengths are related to academic subjects, they do not necessarily lead to increased immediate academic performance. In contrast, C.A.'s interests and strengths greatly contribute to his well-being and self-esteem as a teenager. He connects with his peer and family through his interest in videogames, rap videos, movies, and travel. His strength in music allows him to play in a marching band and perform publicly, which is rewarding for him both socially and because it makes his parents proud, while his calendar calculation abilities impresses others and makes him feel unique. These interests and strengths could therefore further translate into skills or knowledge increasing his quality of life, even if it may not directly translate into better adaptive behaviors or employment (Winter-Messiers, 2007; Davey, 2020).

What Can We Learn From This Case Study?

How can one harness the potential positive effect of interests and relative and absolute strengths? Interests and strengths can contribute to quality of life. They are associated with the experience of positive emotions (Sasson et al., 2012) and can positively affect self-esteem (Chiodo et al., 2017; Happé, 2018) even without an explicit attempt of increasing knowledge of related skills or being a pathway to employment. For example, mood and intrinsic motivation is enhanced when autistic people approach their area of interest (Sasson et al., 2012) and this could be useful for interventions (Davey, 2020). Concerning strengths, their level is independent from their direct usefulness for adaptive outcomes, such as academic performance or paid employment. Promoting the inclusion of interests at home and at school should therefore be cautiously distinguished from expectations regarding the level attained in those domains of interests.

The domains of relative and/or absolute strengths, as well as the domains of deficits, can also be useful for intervention or education by informing which tools or teaching methods are more or less suited for the individual. They can be informative as to what domain or type of material is particularly suited to promote learning for the person. Such strengths may or may not immediately, or ever, lead to a better adaptive outcome, but may contribute to acquiring new skills, hence enhancing general adaptation by benefiting self-esteem, well-being, and mental health. For C.A., being able to pursue his academic curriculum at his own pace, despite the fact that this might not lead to his obtention of a high school diploma, corresponds to his interest and contributes to his self-esteem and feeling of inclusion in society, as he is doing something similar to his peers. Hence, pursuing his education in an adapted college program or finding customized employment which would build on his strengths, could be promising avenues for promoting well-being and quality of life in general.

In conclusion, interests and strengths in autism may not directly lead to an instrumental outcome, such as obtaining paid employment, despite their personally rewarding value and societal utility. For example, the person who has written the most validated Wikipedia articles is autistic[1]. Thus, provided that he is given access to opportunities and becomes an empowered citizen,

[1]https://xtools.wmflabs.org/pages/fr.wikipedia.org/Tsaag%20Valren

the possibilities are endless for 16 year-old C.A. to live a fulfilling life and contribute to society through a paid job or not.

Limitations

This case study has several limitations. We did not have access to the test protocols for the assessments C.A. completed prior to his participation in this study. Such information would have allowed us to better characterize his early cognitive profile and how his strengths and weaknesses evolved with time. More information regarding language development in the three languages he was exposed to would also have been useful to better characterize how this impacted his development in general and hyperlexia in particular, as it was shown that bilingualism can have positive impact on certain cognitive tasks (Trelles and Castro, 2019). Further, no strength-oriented assessment was conducted during preschool or school age. A valid assessment of non-verbal abilities using tests such as the Raven's Progressive Matrices or a formal evaluation of hyperlexia would have been useful to document these early strengths. We were hence limited to infer those strengths from parental reports and qualitative information included in previous reports.

Second, the assessment conducted in the present study was also incomplete in that it did not include many mathematic subtests, nor many music tests. It is possible that some additional strengths in those areas would have become apparent through further assessments, although these turned out to be very challenging and distressing for C.A.. The assessment did not include a direct assessment of distress for C.A. (anxiety or depression scale), which prevented us from doing a pre-post comparison on such measures. It was also relatively short term (14 months).

Third, the measures and intervention mostly relied on C.A.'s mother, as she was more available and hence participated in all assessment and intervention sessions, while the father only attended a few. Finally, it is limited by its design as this is a single case study used to illustrate and discuss how interests and strengths in autism can be harnessed to avoid potential detrimental effects to well-being and quality of life. More examples of both beneficial and detrimental effects of using interests and strengths in intervention in autism are needed.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the CER CIUSSS-NIM. Written informed consent was obtained from the individual(s) legal guardian/next of kin for the publication of the present case-study including anonymized images or data.

AUTHOR CONTRIBUTIONS

VC and LM contributed to the study design, data collection and analysis, intervention with the family, and writing and revisions of the manuscript. VL contributed to the study design, data collection and analysis, intervention with the family, and revisions of the manuscript. PG contributed to the data collection and analysis, intervention with the family, and revisions of the manuscript. AS-D contributed to data collection and analysis and revisions of the manuscript. LB contributed to the study design, to the data collection and analysis, and to the manuscript revisions. AO contributed to the study design and manuscript revision. All authors contributed to the article and approved the submitted version.

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Visual Search for Circumscribed Interests in Autism Is Similar to That of Neurotypical Individuals

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Intense interests are a core symptom of autism spectrum disorders (ASD) and can be all-encompassing for affected individuals. This observation raises the hypothesis that intense interests in ASD are related to pervasive changes in visual processing for objects within that category, including visual search. We assayed visual processing with two novel tasks, targeting category search and exemplar search. For each task, three kinds of stimuli were used: faces, houses, and images personalized to each participant's interest. 25 children and adults with ASD were compared to 25 neurotypical (NT) children and adults. We found no differences in either visual search task between ASD and NT controls for interests. Thus, pervasive alterations in perception are not likely to account for ASD behavioral symptoms.

Keywords: autism spectrum disorder, visual processing, serial processing, parallel processing, circumscribed interests, visual search

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INTRODUCTION

Intense interests are a common symptom of autism spectrum disorder (ASD) (South et al., 2005; Turner-Brown et al., 2011; Klin et al., 2013) and are a specific kind of Restricted and Repetitive Behavior (RRB) (American Psychiatric Association, 2013). The current study focuses on the possible relationship between intense interests and visual processing. Interests are highly motivating for individuals with ASD (Winter-Messiers et al., 2008), and when incorporated into therapy, interests can have a positive effect on ASD clinical outcomes and academic achievement (Boyd et al., 2007; Koegel et al., 2012, 2013; Kryzak et al., 2013; Gunn and Delafield-Butt, 2015; Harrop et al., 2019). However, interests can also be detrimental to daily functioning by interfering in day-to-day activities and social interactions (Klin et al., 2013).

One possible link between interests and visual processing is that ASD symptoms associated with intense interests may produce abnormal visual perception for images related to interests, similarly to how experts demonstrate enhanced visual pocessing for their category of expertise (Gauthier et al., 2000). Alternatively, individuals with ASD may have a primary underlying alteration in the visual system, which leads to intense interests. For example, while neurotypical (NT) controls are hardwired to rapidly process faces and quickly search for faces (Bruce and Humphreys, 1994; Tong and Nakayama, 1999), it is possible that individuals with ASD may respond more quickly to intense interests and show visual expertise for interests similar to how NT controls process faces. We explore these possibilities to better understand the phenomenon of intense interests in ASD.

In order to understand the possible mechanisms of visual expertise for intense interests in ASD, it is important to provide an overview of the forms that visual expertise can take in typical development. For example, visual expertise for faces is widely studied. Face-to-face interactions are the foundation of daily functioning and it is thought that starting early in life, neurotypical individuals are particularly attuned to faces (Mondloch et al., 1999). Evidence for visual expertise for faces comes from a robust behavioral literature (Treisman and Gelade, 1980; Schwarzer, 2000; Hershler and Hochstein, 2005) as well as from functional MRI (fMRI) work, and is supported by neurophysiologic studies in non-human primates (Tsao et al., 2006). Faces uniquely activate a distributed network in the brain that includes the fusiform gyrus (FFA) (Kanwisher et al., 1997; O'Toole et al., 2005), as well as other visual processing areas, including the occipital face area (Anderson et al., 2000).

While visual expertise for faces is pervasive, visual expertise for classes other than faces may also be present in neurotypical individuals (Wood, 1999). For example, a hallmark study by Chase and Simon (1973) demonstrated that chess experts are better at remembering structured chessboard arrangements than novices. More recent eye-tracking studies have shown chess experts make fewer and more holistic fixations when looking at non-random chess board arrangements (Reingold et al., 2001). Visual expertise can also be developed for individuals who spend several hours a day playing hockey (Canal Bruland et al., 2010), video games (Latham et al., 2013), or badminton (Abernethy and Russell, 1987), as well as in certain occupational fields such as medical diagnostics (Crowley et al., 2003) and air traffic control (Van Meeuwen et al., 2014). In a laboratory setting, visual experts have improved short-term memory for their object of expertise (Curby et al., 2009) and have higher signal detection scores (d-prime) when matching different images of the same exemplar object (for example, matching car models from different years) (Gauthier et al., 2000). In all of the above circumstances, individuals demonstrate enhanced visual search and selective attention for their (non-social) expertise.

Visual processing studies in ASD have shown perceptual differences for both social stimuli (faces) as well as non-social stimuli (objects), with some evidence that perception of non-social stimuli in ASD can resemble perception for social stimuli in an NT population (Sasson et al., 2008). Individuals with ASD prefer to look at objects over faces and look at faces less than NT controls (Unruh et al., 2016). These preferences for non-social objects may be present in children diagnosed with ASD as young as two (Klin et al., 2009). Lastly, fMRI studies have demonstrated that individuals with ASD recruit the FFA for non-social objects of interest more than NT controls (Foss-Feig et al., 2016), suggesting that individuals with ASD process interests similarly to how NT individuals process faces. There is a large literature around early visual processing in ASD (Dakin and Frith, 2005; Van der Hallen et al., 2015), with conflicting results depending on what aspect of visual processing is probed. Studies of early visual processing in ASD show enhanced visual processing for fine details, both during visual search (O'Riordan et al., 2001) and in luminance contrast (Luc et al., 2011), but also find deficits in other areas, such as binocular rivalry (Robertson et al., 2013), mental imagery (Marothi et al., 2019), and motion perception (Milne et al., 2002; Bertone et al., 2003), with some work demonstrating this deficit can be found as early as the primary visual cortex (Robertson et al., 2014).

Visual search is a specific type of visual processing that is closely tied to spatial attention (Wolfe, 2015). Visual search tasks involve locating a target item amongst a set of distractor items of variable set size. Visual search is also flexible, with adjusted strategies based on set size and complexity (Wolfe et al., 1992) and separable from working memory (Horowitz and Wolfe, 1998). In NT individuals, visual search tasks involving faces demonstrate high efficiency in search compared to other object types (Bruce, 1986), even for faces that are only viewed for a brief period of time (Diamond and Carey, 1986).

In one common visual search paradigm, participants must have *categorical* knowledge of an object in a specific category, or knowledge about how the object is different from objects in other categories. In this paradigm, participants search for images of a particular category (butterflies or cars, for example) amongst an array of unrelated distractor images, such as animals or articles of clothing. Experts in a particular category have higher search efficiency on that category than non-experts (Hershler and Hochstein, 2009; Golan et al., 2014). In a contrasting type of visual search paradigm, participants must have *exemplar* knowledge, meaning that they must be able to pick out an image that is consistent with a category of distractor images. For the bird category of the Vanderbilt Expertise Test, for example, participants spend several seconds viewing a group of images of birds, followed by a second set of novel bird images in which the participant must find an image that depicts a matching species from the first group (McGugin et al., 2012). These two paradigms differ in the distinction (category vs. exemplar) that must be picked out during visual search. Furthermore, category and exemplar search differ in complexity and difficulty, with category search requiring the knowledge of early visual components of a category, and exemplar search requiring broader knowledge about specific instantiations of a category.

As visual expertise is not as a monolithic process, consideration must be given to the origins of alterations in the visual pathway. There are two overall ways that visual expertise and intense interests may be related in ASD: intense, non-social interests may alter visual experience, leading to expertise, or alterations in the normal development of the visual system may result in object categories taking over circuitry that is typically specialized for faces, leading to the development of intense, non-social interests. Given the alterations of spatial attention in ASD (Townsend et al., 2001; Sokhadze et al., 2016), visual search is a particularly relevant method for understanding visual processing in ASD. Visual search tasks readily measure certain aspects of visual expertise, and can test whether intense interests are indeed associated with a shift in this domain. Prior work on visual search in ASD suggests enhanced visual search abilities with neutral object stimuli such as shapes, letters, or common objects, as compared to NT controls, with faster reaction times and higher accuracy levels (Joseph et al., 2009; Kaldy et al., 2016). It is unknown whether individuals with

ASD will demonstrate enhanced visual search capabilities for individualized interests.

The present study tested visual expertise for intense interests in children and adults with ASD compared to controls with two novel visual search paradigms that distinguished category vs. exemplar search abilities (Jonides and Gleitman, 1972; Smilek et al., 2006). Building upon prior visual expertise paradigms, personalized images of each participant's interest or hobby were compared to images of faces and houses. Given the work that demonstrates that non-social objects are processed atypically in ASD, and that categories of expertise can be accompanied by enhanced visual search abilities, we hypothesized that visual search abilities to intense interests in ASD would be enhanced in both the category and exemplar tasks, resulting in reduced reaction times or possibly greater search efficiency. Enhanced performance in either of these tasks would suggest that intense interests in ASD are a visual atypicality. Inclusion of both a category and an exemplar task, which draw on different visual search processes, allowed us to be more specific in our diagnosis of the origin of intense interests and to increase our ability to identify a visual-based performance difference. We also predicted enhanced visual search skills for faces in NT controls would not be observed in individuals with ASD.

Finally, we mention another advantage of studying search: as mentioned above, in NT subjects, search tasks involving faces are substantially more efficient than search tasks for other object categories (Bruce, 1986; Diamond and Carey, 1986). As this is a robust and consistent finding in NT subjects, we reasoned (and the statistical analyses below confirm) that a modest subject pool has high power in identifying whether this characteristic of search is substantially altered in ASD subjects.

MATERIALS AND METHODS

Participants

Fifty participants (children ages 5–16 and adults ages 18–30) completed one of two tasks–a category search task and an exemplar search task, both described in detail below. 32 participants (17 ASD, 18 children) completed the category search task and 30 participants (16 ASD, 18 children) completed the exemplar search task; 12 participants completed both–eight ASD (five children, three adults) and four NT (four children, zero adults). Two of the 17 ASD participants who completed the category task were excluded from analyses due to incomplete data. Of the adults with ASD, five were their own legal guardian, and four had a caregiver as their guardian. Of the children with ASD, all attended school full-time. Participants with ASD (six females) were recruited through the Center for Autism and the Developing Brain (CADB) in White Plains, NY, United States. Neurotypical (NT) controls (nine females) were recruited through the Sackler Institute for Developmental Psychobiology in Manhattan, NY and through the local New York City community. Informed written consent (assent from minors, consent from caregivers) was obtained from all participants and the study protocol was approved by the Weill Cornell Medicine Institutional Review Board.

Phone Interview

One to two weeks before a participant's in-person testing, a 5-min telephone interview was conducted to assess participants' primary interests. For participants under 13 years old, the interview was conducted with a caregiver. Only two participants in the child group did not fall into this category. The participant (or caregiver) was asked to name three activities or topics that he or she enjoyed doing or thinking/learning/talking about. For each interest, the participant was asked to elaborate on specific aspects of the interest that he or she liked, to indicate how long he or she has had this interest, and whether the interest had changed or developed over time. The participant was also asked to specify which of the three interests were most prominent at the time of the interview. The questions were designed to target the specific aspects of the topic or activity that was most appealing in order to identify stimuli to be used in the tasks. Multiple interests were queried in case the most prominent interest could not be easily represented visually (such as listening to music). ASD participants and caregivers consistently reported interests that were more intense and more specific (as indicated by statements such as "he watches the same movie over and over again," or a preference for particular movies or episodes in a series as opposed to the series as a whole) than those reported by NT caregivers. All answers were recorded on paper and stored with the participant's data folder.

Autism Assessments and Cognitive Testing

Participants with ASD received a diagnosis from a trained clinician at CADB using Module three or four of the Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2012) prior to participation. Total calibrated severity scores (CSS) were generated from the ADOS as well as for Social Affect (SA) and RRB (Hus et al., 2014). NT participants under 18 years old were screened for ASD symptoms with the Social Communication Questionnaire (SCQ-Lifetime) (Rutter et al., 2003), and participants 18 years old and older were screened with the Autism Spectrum Quotient (AQ) (Baron-Cohen et al., 2001). Participants were deemed eligible if they had scores under 15 on the SCQ and scores under 32 on the AQ. Two participants were missing SCQ scores, and in these cases the Social Responsiveness Scale-2 (SRS-2) (Constantino and Gruber, 2012) was used, with a cutoff score of 70. One NT participant was excluded from category task analyses based on their SCQ score. Cognitive skills were measured in participants under 16 years of age with the Differential Abilities Scale-II (school age) (DAS) (Elliott, 2007), and participants 16 years old and older completed the Wechsler Adult Intelligence Scale (WAIS-IV) (Wechsler, 2008). Standard scores for verbal IQ (VIQ) and non-verbal IQ (NVIQ) were derived from the DAS-II or WAIS-IV (see **Table 1** for full demographic information).

Interest Assessments

At the in-person visit, participants (or caregivers) completed a questionnaire about the participant's topic or activity of interest identified through the phone interview. There was a child version

TABLE 1 | Participant demographics.

	ASD Children		ASD Adults		NT Children		NT Adults	
	Category	Exemplar	Category	Exemplar	Category	Exemplar	Category	Exemplar
Age–Mean (Range)	10.73 (5.75–15.83)	12.09 (7–16.17)	25.53 (20.08–30.33)	24.96 (21.33–31.25)	9.92 (6.25–12)	9.63 (6.08–13.08)	22.58 (19.83–27.67)	25.15 (21.58–29.33)
# of Females/Males	2/9	1/9	4/2	3/3	2/8	1/7	2/3	4/2
AQ–Mean (SD); Range	N/A	N/A	N/A	N/A	N/A	N/A	13.2 (4.09); 7–18	22.5 (3.62); 18–27
SCQ–Mean (SD); Range	N/A	N/A	N/A	N/A	5.13 (5.33); 1–16	4.13 (3.31); 1–9	N/A	N/A
VIQ–Mean (SD); Range	98.81 (19.5); 71–143	103.10 (20); 71–143	104 (8.74); 95–118	101 (5.33); 95–108	112.30 (14.2); 90–136	115.75 (17); 86–136	124.20 (13.7); 110–145	113.17 (10.5); 102–127
NVIQ–Mean (SD); Range	96.81 (15.3); 77–131	101.30 (13.6); 84–131	98.5 (13.5); 81–119	95.83 (8.64); 81–104	108.80 (18.2); 89–149	108.13 (14.3); 80–121	116.60 (8.65); 105–127	106.67 (6.98); 98–117
ADOS CSS–Mean (SD); Range	7.20 (1.99); 4–10	7.50 (1.96); 4–10	7.67 (0.816); 7–9	8 (0.894); 7–9	N/A	N/A	N/A	N/A

administered to caregivers and an adult version completed via self-report. The questionnaire asked the caregiver or participant to specify what they knew about or did involving their topic or activity of interest, how much it interfered with day-to-day activities such as spending time with friends/family and going to school/work, and to indicate the duration of their interest on a 1–5 scale (1 = less than 6 months, 5 = over 5 years). From the questionnaire, two scores were derived: an "Interference" measurement, defined as the average rating on the questions concerned with how much the interest took time from activities related to friends, family, school and/or work, and a "Current Time" measurement, defined as the average rating on questions concerned with the amount of time spent on the interest on a day-to-day basis. On the child version, scores ranged from 1 to 3 (less than 25% of the time, 25–75% of the time, over 75% of the time), and on the adult version, scores ranged from 1 to 5 (1 = strongly disagree, 3 = neither agree nor disagree, 5 = strongly agree), and were converted to a one to three scale to match the child version. While using two different questionnaires may make it more difficult to compare scores, each version of the questionnaire was designed to be completed by a specific age range, and thus differentiating them was necessary.

Category Search Task

This task, presented on an iPad (Model number: A1822, 9.4 in. × 6.6 in.), made use of three categories: Houses, Faces, and Interests (**Figure 1A**). Stimuli for Houses were 108 unique photos of houses gathered from the internet and a stimulus set by Konkle et al. (2010). Stimuli for Faces were 108 unique full-face photos of child and adult faces from the Developmental Emotional Faces Stimulus Set by Meuwissen et al. (2017). While this stimulus set has not been previously used in an ASD population, it was chosen because the age range of faces (8–30 years old) was similar to the age range of the participants. To avoid possible confounds due to differences in emotional processing between NT and ASD individuals, only happy faces were used. The Interests category was individually tailored for each participant based on the phone interview; for example, a participant who indicated on the phone that his/her primary interest was the video game "Minecraft" saw screenshots from the video game (see **Figure 1A**). Interests stimuli were 108 unique photos of the participant's interest gathered from the internet (see **Supplementary Table S1** for a list of interests). While some of the Interests stimuli were related to people, such as TV shows or movies, and therefore contained faces, none of the images displayed faces in a prominent manner, thus distinguishing them from the large, centered, and in-focus faces in the Faces condition. All stimuli were resized to 256 × 256 pixels using MATLAB software.

There were three practice trials and 108 test trials per category. A trial consisted of either 4, 16, or 36 images in a random array for 2,000 ms, followed by a central fixation cross for 1,000 ms. The trial duration of 2,000 ms was used based on the performance of pilot subjects. There were 36 trials for each array size. In each trial, images were presented in a random array (see **Figure 1B**). One image, the target, was intact; the distractor images were created by scrambling the target image based on a random repositioning of an 8 × 8 grid of sub-blocks. Scrambled images were used as

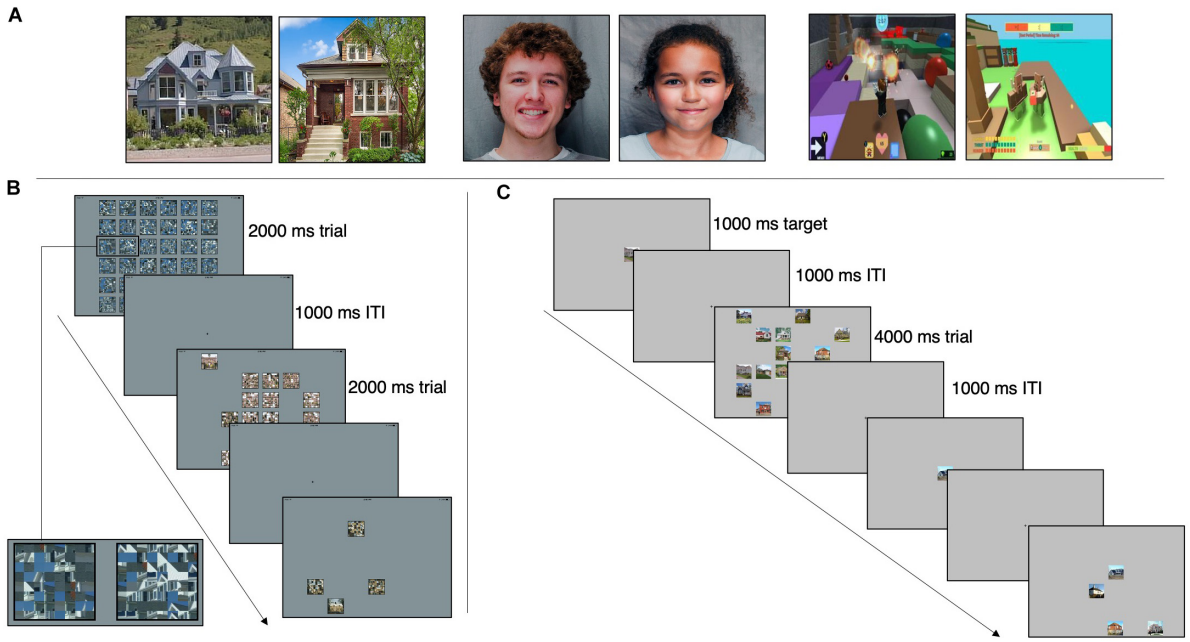

FIGURE 1 | (A) Example stimuli from the houses, faces, and interests categories (left to right). House stimuli are from Konkle et al. (2010) and face stimuli are from Meuwissen et al. (2017). **(B)** Category Search Task. Participants instructed to find the unscrambled image. Three example trials displayed. 36-image array presented for 2,000 ms with 35 scrambled images and one target, a 1,000-ms ITI, followed by a 16-image array and ITI, and lastly a four-image array. Box in first image of sequence is enlarged to show example of scrambled images. **(C)** Exemplar Search Task. Participants instructed to find the target image. Two example trials displayed. Target presented for 1,000 ms, a 1,000-ms ITI and a 16-image array with 15 distractors and the target image. A 1,000-ms ITI separates this trial from the next target presentation, which is part of a four-image array trial.

distractors as opposed to other-category images (as used in Golan et al., 2014 and Hershler and Hochstein, 2009) to avoid potential visual processing differences in ASD for different categories of objects, which could have confounded interpretation of a positive result. In addition, some categories of objects are more similar than others; scrambling images allowed the distractor difficulty to be standardized. The same image size was used for all three array sizes. A constant image size but variable array size was chosen so that the slope of RT vs. array size could be measured without a size confound. This is a standard approach in studies of visual search (Tong and Nakayama, 1999; Smilek et al., 2006). Images were scrambled using MATLAB.

Participants were instructed to find and touch the target as quickly as possible. The position of the target in the array was randomized, but the average target position across all trials was the center of the array. Trial order was randomized, and blocks for each category were run in random order. Participant accuracy (correct or incorrect) and reaction times were recorded for each trial.

Exemplar Search Task

The exemplar task, presented on the same iPad from the category task, consisted of the same three categories (Houses, Faces, and Interests) as the category task. Image size was the same as in the category task. Each category contained 54 trials. A trial consisted of a single target image presented at the center of the display for 1,000 ms, a 1,000 ms crosshair, and then the target and either 3, 8, or 15 distractors in a random array for 4,000 ms (a longer search time than that of the category task due to the increased difficulty of this task). In contrast to the category search task, distractor images were not scrambled images but were different examples drawn from the same category as the target (see **Figure 1C**). There were 18 trials for each array size. Participants were instructed to find and touch the target as quickly as possible. Each trial's target was unique, but the distractors repeated between trials. Trial order and category order were randomized. Participant accuracy (correct or incorrect) and reaction times were recorded for each trial.

Data Analysis

Primary analyses were identical for both tasks. Accuracies and average reaction times (RTs) were calculated for each array size and category. Accuracy was defined as number of correct trials out of the total number of trials for each array size. For each category, a slope (milliseconds/item) was calculated from the average RTs, determined from the regression (least-squares) of average RT vs. array size. Trials in which no response was registered in the allotted time (2,000 ms for the category task and 4,000 ms for the exemplar task) were counted as misses in the accuracy measurement and were excluded from all RT analyses.

To assess the effects on accuracy and RTs, a 3 (category) × 3 (array size) × 2 (diagnosis) × 2 (age group) ANOVA was performed for both measures. To assess the effects on slope, a 3 (category) × 2 (diagnosis) × 2 (age group) ANOVA was

performed. Age of participants was binarized into two groups, children and adults. *p*-values from the ANOVA are reported without correction for multiple comparisons, as our main focus is on whether there is an interaction between diagnosis and category (a single comparison for each ANOVA), and we wanted to maximize the sensitivity to detect such interactions. Significant main effects and interactions were interrogated with *post hoc* *t*-tests. In the body of the "Results" section, the *F*-values and *p*-values are provided for significant effects and interactions, and only the *p*-value is provided for non-significant effects and interactions. The full statistics for all tests can be found in **Tables 2**, **3**.

RESULTS

Questionnaires

On the interest questionnaire, adults with ASD scored higher on Current Time than NT adults ($t(15) = 3.972$, $p = 0.001$), but there was no difference in Interference ($t(15) = -0.763$, $p = 0.458$). In the child group, there was a trend of a difference in Interference ($t(26) = 1.887$, $p = 0.070$), but no difference in Current Time ($t(26) = 1.587$, $p = 0.125$), likely due to the limited range in response options on the child version compared with the adult version.

Verbal and non-verbal IQ scores were significantly different or nearly so for both the category task and the exemplar task (VIQ category task: $t(30) = -2.822$, $p = 0.008$; NVIQ category task: $t(30) = -2.633$, $p = 0.013$; VIQ exemplar task: $t(28) = -2.242$, $p = 0.033$; NVIQ category task: $t(28) = -1.929$, $p = 0.064$), with ASD participants demonstrating lower scores than NT participants. However, with age and diagnosis as regressors, neither slope, accuracy, nor RT were significantly correlated with IQs on either the category task (p's > 0.318) or the exemplar task (p's > 0.088).

There was a significant difference between AQ scores of participants in the category task vs. the exemplar task ($t(9) = -4.006$, $p = 0.003$).

Category Task

Accuracy

Overall accuracy on the task was high, on average 92%. Accuracy for Faces was highest, followed by Houses and Interests (see **Table 2** for full statistics, including *F*-values for non-significant comparisons). As expected, accuracy was highest for the smallest array size, and decreased as array size increased. There was a trend of a main effect of age with adults having an overall higher accuracy than children (Adults Mean: 96%; Children Mean: 90%, $p = 0.08$) (see **Figure 2**). However, there was no main effect of diagnosis on accuracy ($p = 0.745$), and no interaction between diagnosis and category ($p = 0.382$).

Reaction Time

Participants' reaction times were different for each category ($F(2,50) = 156.534$, $p < 0.001$) with faster RTs for Faces than for Houses and for Interests, and faster RTs for Houses than for Interests. Reaction times were also influenced by array size ($F(2,50) = 149.330$, $p < 0.001$) with faster RTs for smaller array sizes. An interaction between array size and category ($F(4,100) = 26.669$, $p < 0.001$) was explained by less change in RTs for Faces across array size compared to Houses and Interests.

Adults had faster RTs than children ($F(1,25) = 7.907$, $p = 0.009$). An interaction between array size and age ($F(2,50) = 7.504$, $p = 0.001$) was explained by a larger gap in RTs between adults and children on smaller array sizes than on larger ones (see **Figure 2**). However, there was no significant main effect of diagnosis on RTs ($p = 0.290$), and no interaction between diagnosis and category ($p = 0.709$).

Slope

Slope changed with category ($F(2,50) = 44.520$, $p < 0.001$) as participants had lower slopes for Faces relative to Houses and Interests. There was no difference in slope between Houses and Interests.

There was a main effect of age ($F(1,25) = 12.647$, $p = 0.002$). While adults overall had lower RTs than children (see above), adults overall had higher slopes than children (see **Figure 2**). Given that the slope is a value derived from the average RT values for each array size, this suggests that on Houses and Interests, while children performed worse than adults on smaller array sizes, as array size grew the age-related performance gap shrunk. There was no effect of diagnosis ($p = 0.611$) on slope, and no interaction between diagnosis and category ($p = 0.929$).

Exemplar Task

Accuracy

Overall accuracy on the task was 79%. Accuracy was impacted by category ($F(2,50) = 12.598$, $p < 0.001$) and was higher for Interests than for Faces, and was higher for Faces than for Houses (see **Table 3** for full statistics). As expected, array size impacted accuracy ($F(2,50) = 73.139$, $p < 0.001$), with a decrease in accuracy as array size grew.

Adults had higher accuracy than children ($F(1,25) = 11.097$, $p = 0.003$). While accuracy for some children was quite low (below 60%), all participants exhibited the same decrease in accuracy as array size increased, suggesting that the low accuracy was a result of an overall increase in task difficulty, rather than a misunderstanding of task instructions (see **Figure 3**). There was no effect of diagnosis on accuracy ($p = 0.895$), and there was no interaction between diagnosis and category ($p = 0.550$).

Reaction Time

Category did not impact RTs ($p = 0.169$). RTs were impacted by array size ($F(2,50) = 114.863$, $p < 0.001$), with RTs increasing as array size increased.

Adults had lower RTs than children ($F(1,25) = 13.42$, $p = 0.001$) (see **Figure 3**). There was no significant effect of diagnosis ($p = 0.380$) and no significant interaction between diagnosis and category ($p = 0.894$).

Slope

Category ($p = 0.230$) and age ($p = 0.280$) did not influence slopes (see **Figure 3**). The lack of difference in age, paired with the distinct differences in age on RTs and accuracies,

TABLE 2A | Category task statistics.

		Accuracy		Reaction Time		Slope		
		F	*P*	*F*	*p*	*F*	*p*	DoF
Main Effects	Dx	0.108	0.745	1.168	0.290	0.265	0.611	(1, 25)
	Age	3.334	0.080	7.907	0.009	12.647	0.002	(1, 25)
	Category	12.678	<0.001	156.534	<0.001	44.520	<0.001	(2,50)
	Array Size	55.565	<0.001	149.330	<0.001			(2, 50)
Two-Way Interactions	Dx × Age	0.075	0.787	0.979	0.332	1.092	0.306	(1, 25)
	Dx × Category	0.980	0.382	0.347	0.709	0.074	0.929	(2, 50)
	Age × Category	2.492	0.002	0.250	0.780	12.533	<0.001	(2,50)
	Dx × Array Size	0.736	0.484	0.547	0.582			(2, 50)
	Age × Array Size	3.594	0.035	7.504	0.001			(2, 50)
	Category × Array Size	25.836	<0.001	26.669	<0.001			(4, 100)
Three-Way Interactions	Dx × Age × Category	0.135	0.874	1.099	0.341	0.361	0.699	(2, 50)
	Dx × Age × Array Size	1.103	0.340	0.642	0.430			(2, 50)
	Dx × Category × Array Size	0.187	0.945	0.472	0.757			(4, 100)
	Age × Category × Array Size	0.794	0.532	7.719	<0.001			(4, 100)

Dx, Diagnosis; DoF, Degrees of Freedom.

TABLE 2B | Category task statistics (*Post hoc* analyses).

		Accuracy		Reaction Time		Slope	
	Comparison	*T*	*p*	*t*	*p*	*t*	*p*
Category	Faces vs. Houses	3.401	0.002	−14.313	<0.001	−6.658	<0.001
	Faces vs. Interests	6.713	<0.001	−18.217	<0.001	−5.437	<0.001
	Houses vs. Interests	1.269	0.215	−4.108	<0.001	−0.154	0.879
Array Size	4 vs. 16	2.313	0.028	−3.133	0.004		
	4 vs. 36	8.220	<0.001	−12.768	<0.001		
	16 vs. 36	10.091	<0.001	−11.257	<0.001		

Degrees of Freedom for all comparisons = 28.

TABLE 3A | Exemplar task statistics.

		Accuracy		Reaction Time		Slope		
		F	*P*	*F*	*p*	*F*	*p*	DoF
Main Effects	Dx	0.018	0.895	0.811	0.376	0.352	0.558	(1, 25)
	Age	11.097	0.003	13.420	<0.001	1.229	0.278	(1, 25)
	Category	12.598	<0.001	1.839	0.169	1.503	0.232	(2, 50)
	Array Size	73.139	<0.001	114.863	<0.001			(2, 50)
Two-Way Interactions	Dx × Age	1.245	0.275	2.702	0.113	0.021	0.885	(1, 25)
	Dx × Category	0.606	0.550	0.113	0.894	0.105	0.900	(2, 50)
	Age × Category	1.025	0.366	1.315	0.278	0.185	0.832	(2, 50)
	Dx × Array Size	0.136	0.873	1.210	0.307			(2, 50)
	Age × Array Size	3.717	0.031	1.247	0.296			(2, 50)
	Category × Array Size	2.488	0.048	1.174	0.327			(4, 100)
Three-Way Interactions	Dx × Age × Category	0.699	0.502	0.603	0.551	0.003	0.997	(2, 50)
	Dx × Age × Array Size	1.782	0.179	0.016	0.984			(2, 50)
	Dx × Category × Array Size	1.134	0.345	0.092	0.985			(4, 100)
	Age × Category × Array Size	0.554	0.697	0.288	0.885			(4, 100)

Dx: Diagnosis; DoF: Degrees of Freedom.

TABLE 3B | Exemplar task statistics (*Post hoc* analyses).

		Accuracy		Reaction Time		Slope	
	Comparison	***t***	***P***	***T***	***p***	***t***	***p***
Category	Faces vs. Houses	2.392	0.024	0.532	0.599	0.691	0.495
	Interests vs. Houses	5.002	<0.001	−1.391	0.175	−1.089	0.285
	Interests vs. Faces	3.338	0.002	−2.124	0.042	−1.971	0.058
Array Size	4 vs. 9	4.121	<0.001	−14.300	<0.001		
	4 vs. 16	9.864	<0.001	−13.225	<0.001		
	9 vs. 16	9.857	<0.001	−6.334	<0.001		

Degrees of Freedom for all comparisons = 28.

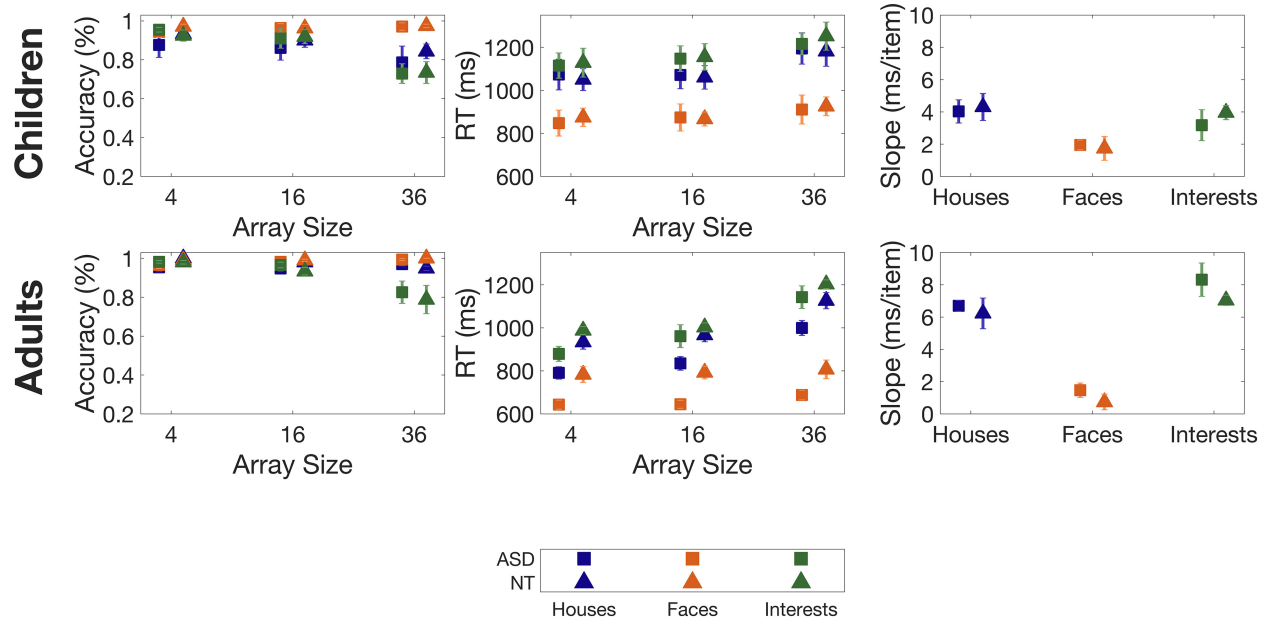

FIGURE 2 | Accuracies, Reaction Times (RTs) and slopes for the category task, for children (top panels) and adults (bottom panels). Slopes are determined from a least-squares regression of RT vs. array size. Error bars indicate 1 Standard Error of the Mean.

suggests that while children performed worse on the task than adults overall, both groups were affected by the increase in array sizes equally. There was also no main effect of diagnosis ($p = 0.558$), and there was no interaction between diagnosis and category ($p = 0.900$).

Power Analyses

As our findings did not reveal a significant difference in search performance in ASD participants vs. NT controls, we undertook power analyses to determine the likelihood that, if substantial differences were present, they would have been detected. Power analyses are summarized in **Table 4** and detailed below. Briefly, owing to the consistency of findings in NT subjects, the category task has power of >98% in revealing either an absence of a greater efficiency for Faces, or a reversal of efficiency between Faces and Interests. The exemplar task was underpowered for identifying an absence of differential efficiency for Faces (17%), and had a power of approximately 70% for revealing a reversal, but nevertheless adds to the overall power of the study.

The power analyses were conducted via a bootstrap, a standard procedure for determining study power *post hoc* (Efron and Tibshirani, 1998; Walters, 2004). We considered two hypothetical scenarios in which the well-known specialized processing for faces expected in NT subjects (and confirmed here) might be altered in a way that could account for ASD symptomatology. In scenario (i), individuals with ASD lacked the difference in efficiency for Faces compared to Interests as seen in NT participants (greater in the category task, lesser in the exemplar task), and instead processed Faces and Interests in the same way. In scenario (ii), individuals with ASD showed the reverse of the pattern seen in NT participants; for the category task, this means processing Interests more efficiently than Faces, and for the exemplar task, Faces more efficiently than Interests.

For each scenario, the sensitivity was estimated by creating 1,000 surrogate datasets conforming to the hypothesis, and determined how often a significant interaction between diagnosis and category would have been obtained by our analytical procedures. The NT components of the surrogate datasets were

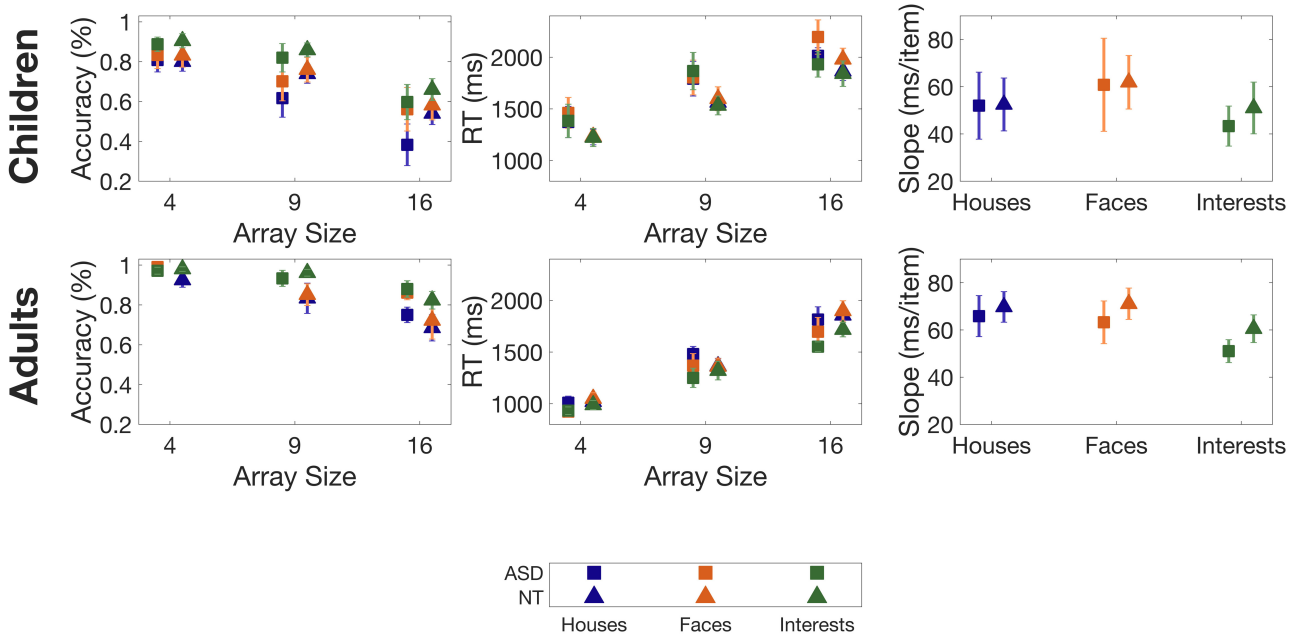

FIGURE 3 | Accuracies, Reaction Times (RTs) and slopes for the exemplar task, for children (top panels) and adults (bottom panels). Slopes are determined from a least-squares regression of RT vs. array size. Error bars indicate 1 Standard Error of the Mean.

TABLE 4A | Power Analysis, scenario (i): no difference between faces and interests in ASD subjects.

		Number of significant ANOVAs out of 1,000			
		Accuracy	**Reaction Time**	**Slope**	**Overall**
Dx × Category ANOVA	Category Task	481	956	376	987
	Exemplar Task	76	50	56	171

TABLE 4B | Power Analysis, scenario (ii): reversed difference between faces and interests in ASD subjects.

		Number of significant ANOVAs out of 1,000			
		Accuracy	**Reaction Time**	**Slope**	**Overall**
Dx × Category ANOVA	Category Task	1,000	1,000	999	1,000
	Exemplar Task	581	163	176	714

generated by standard bootstrapping (i.e., random sampling with replacement) from our sample. The ASD components were also generated by bootstrapping, but the data from each participant were modified to simulate each of the above scenarios. Specifically, in scenario (i), the data for the Faces and Interests trials were randomly interchanged; in scenario (ii), they were systematically swapped. Each of these surrogate datasets was then analyzed in the same way as the actual data, with ANOVAs conducted for the three performance measures (Accuracy, Reaction Time, and Slope) in each of the two tasks. A surrogate dataset was considered to yield a positive result if the p-value for the interaction between diagnosis and category was <0.05. Note that, as with the analysis of the actual data, these p-values were not corrected for multiple comparisons.

Table 4 reports the results of this analysis. If ASD participants differed from NT participants by having no difference between Faces and Interests (scenario (i), **Table 4A**), then a significant interaction would be present for at least one of the three performance measures in 987/1,000 of the surrogate datasets in the category task. If the difference between Faces and Interests were reversed (scenario (ii), **Table 4B**), then a significant interaction would be present for at least one of the three measures in all of the surrogates (1,000/1,000) in the category task. Reaction time was the most sensitive of the three measures. The exemplar task was less sensitive to detecting these two scenarios [171/1,000 for scenario (i), 714/1,000 for scenario (ii)], with accuracy being the most sensitive indicator.

These analyses also provide estimates of power for scenarios in which ASD subjects have a greater performance difference

for Interests than Faces in the exemplar task, compared to NT subjects. This is because (as is standard) the ANOVA assumes that main effects are additive, and interactions are multiplicative. Thus, the analysis of **Table 4A**, in which the interaction is equal to the size of the Interests vs. Faces difference in NT participants, applies not only when the interaction cancels the Interests vs. Faces difference, but also to the case in which it reinforces this difference (and therefore doubles it). As a result, the sensitivity to detect a doubling of the Interests vs. Faces difference in ASD vs. NT participants in the exemplar task is also given by **Table 4A**, lower row. Similarly, the analysis of **Table 4B**, in which the interaction is double the size of the Interests vs. Faces difference in NT participants, applies not only when it reverses the Interests vs. Faces difference, but also to the case in which it reinforces this difference (and therefore triples it). This means that the sensitivity to detect a tripling of the Interests vs. Faces difference in ASD vs. NT participants in the exemplar task is also given by **Table 4B**, lower row. Note, however, that the data (**Figure 3**) show no suggestion that ASD and NT participants differed in terms of their performance on Interests vs. Faces.

Thus, despite the modest sample size, the category task showed good sensitivity for detecting either of two plausible alterations in the ASD population–likely because the main effect of category was robust ($p < 0.001$ for all three measures). The exemplar task had much lower sensitivity for these specific scenarios, but it could have revealed kinds of differences that the category task overlooked.

DISCUSSION

The present study sought to examine whether ASD individuals demonstrate a visual processing advantage for unique interests compared to NT controls. We tested this using two visual search tasks: category search and exemplar search. These tasks make different demands on visual processing and tap distinct aspects of early visual search skills: basic classification and subordinate classification. In the exemplar task, RTs were longer for larger array sizes, while in the category task, there was little change in RTs with array size, consistent with prior studies of similar tasks (Jonides and Gleitman, 1972; Smilek et al., 2006). Contrary to our hypotheses that intense interests in ASD may lead to, or result from, differences in early stages of visual processing, there was no evidence of differences between the performance of NT controls and ASD individuals for Interests in either task, as well as no differences for Faces and Houses. Neither children nor adults with ASD demonstrated evidence of visual expertise for their interests relative to age-matched NT controls, even though adults with ASD reported spending more time on their interests than NT adults. The findings are similar to prior work demonstrating no differences in attention (Parsons et al., 2017) or learning (Schuetze et al., 2019) for personalized interests in ASD as compared to NT controls, as well as no differences in visual acuity (Tavassoli et al., 2011). Together the findings suggest that while these search tasks captured low-level visual perceptual differences across key variables (i.e., improved performance with age, increasing RT with array size, predominantly parallel processing for category search), differences in low-level visual perception between ASD and NT participants are relatively minor. While the diagnostic differences in this study are a null finding, this does not rule out the possibility of diagnosis-based differences in visual processing at later stages, or that minor differences in perception are present. Instead, the results demonstrate that the processes required for the current tasks are not large enough to account for the diagnostic behavioral discrepancies between ASD and NT individuals regarding faces and intense interests.

If no causative differences for intense interests occur during early visual perception, then perhaps ASD symptoms relating to intense interests are explained by mechanisms later in the processing stream directly related to reward valuation and executive functioning. Our own work, as well as that of others, suggests that interests are particularly motivating for individuals with ASD. When individuals with ASD observe images of interest they demonstrate greater feelings of pleasure (Sasson et al., 2012). In economic choice paradigms, individuals with ASD value their interests more than a group of NT controls (Watson et al., 2015). Further, regions important for processing arousal, such as the anterior insula (Cascio et al., 2014), as well as reward circuitry, including the dorsal striatum (Kohls et al., 2018), were more sensitive to interests in individuals with ASD than NT controls. Our group has demonstrated that images of interest can interfere with cognitive control in children with ASD but not NT controls (Bos et al., 2019). The present findings suggest that it is likely that intense interests interfere with cognition at the level of arousal and cognitive control in ASD but not visual perception. Future research should seek to directly compare the effects of intense interests on visual perception with the effects on cognitive control. A within-subjects design that utilizes tasks that probe both early visual processing as well as executive functioning may reveal when in the processing stream the differences between ASD and NT individuals occurs.

In both tasks, participants' accuracy was impacted by category, with highest accuracy for Faces in the category task and for Interests in the exemplar task. In the exemplar task, there was no impact of category on RTs. Overall, the accuracy findings are consistent with our prediction that participants would respond differently to each category of images (Levin et al., 2001). In the exemplar task, there were longer slope values and lower accuracies than in the category search task. This suggests that participants primarily relied on serial processing strategies for the exemplar task and parallel processing strategies in the category task. These behavior patterns are consistent with prior work suggesting that serial processing relies on slower visual strategies compared to parallel processing (Eriksen and Spencer, 1969; Shiffrin and Gardner, 1972). The highly stereotyped nature of responses for both of these visual search skills precludes the need for a within subject paradigm that directly compares performance between these two types of tasks. It is also possible that the exemplar task also had a working memory component, given the need for participants to remember a particular stimulus after a delay. An enhanced working memory for objects of interest may explain why participants were more accurate for interests than for faces. However, there was no significant difference of this effect

across diagnostic groups, which is in line with past work that demonstrates working memory differences in ASD only at high working memory loads (Steele et al., 2007).

Given that symptomatology and visual expertise varies with age, participants were divided into two age groups in order to ascertain effects of age on task performance. Consistent with prior visual search studies (Kail, 1991; Donnelly et al., 2007), children had longer reaction times and were less accurate than the adult participants. Also as expected, all participants were faster and more accurate for smaller array sizes in both tasks (Kwak et al., 1991). There were no observed interactions between diagnosis, age, and task performance for Faces, Interests, or Houses. Together these results highlight that both tasks successfully captured early visual search perception in children and adults. While NT participants had significantly higher IQs than ASD participants, this is a well-recognized trait difference in ASD (Richler et al., 2007). In addition, neither VIQ nor NVIQ was related to task performance, demonstrating that the current findings cannot be explained by group IQ differences. Finally, while AQ scores were significantly different between tasks, this is unlikely to explain any results, as the pattern of results across the two tasks was highly similar and all participants were under the cut-off of 32, as suggested by Baron-Cohen et al. (2001).

Interestingly, there was no impact of diagnosis on performance for Faces in either task. These findings were surprising given prior work that has shown general differences in visual processing and visual attention for faces in ASD as compared to NT controls (Boucher et al., 1998; Dalton et al., 2005; Uljarevic and Hamilton, 2013). However, some studies have shown that individuals with ASD are similar to NT controls for certain aspects of low-level face configuration processing. For example, individuals with ASD are susceptible to the face inversion effect (Teunisse and De Gelder, 2003) and are able to detect gaze direction at the same level as NT controls (Gepner et al., 1996). The literature is also mixed on the ability of individuals with ASD to detect facial expressions (Jemel et al., 2006). One possibility is that the visual search paradigm, in which individuals with ASD are known to have an advantage (O'Riordan et al., 2001; Simmons et al., 2009; Kaldy et al., 2016), may have masked the typical processing deficiencies for faces that individuals with ASD exhibit. Future work that examines the confound of enhanced visual search abilities in ASD in domains where individuals with ASD are traditionally impaired, such as face processing, would be helpful in understanding these results. Another possibility is that given the significant heterogeneity associated with ASD (Lord and Jones, 2012), the subset of ASD individuals who completed this task had less severe face processing difficulties than other subgroups of individuals on the spectrum.

There were certain limitations to the present study. First, the interest questionnaire used a different scale for children and adults, making it difficult to combine data across age groups. The child version of the questionnaire also had a limited response range, making it challenging to draw conclusions about the nature of the interests. The images of the interests themselves varied in complexity, which could have affected task performance between participants. However, while we do not quantify the level of complexity for each interest, an examination of the interest list for each group does not suggest a difference in image complexity between groups. More importantly, a complexity difference might lead to a spurious performance difference between the groups, not a lack of difference, as we found. Thus, it is unlikely that image complexity affected the central conclusions of the study. Future studies may wish to systematically manipulate image complexity of both targets and distractors.

The scrambled-distractor paradigm in the category task may be substantially easier than other types of category tasks that use other-category distractors. Although it does not seem that there was a ceiling effect, since there were significant differences in slope across categories, as well as noticeable decreases in accuracy and increases in reaction time across array sizes, future studies may wish to compare category task performance with a scrambled-distractor paradigm to performance with an other-category paradigm. Finally, the number of female participants was too small to assess sex effects in the analyses, which may be informative given the sex imbalance in ASD and the possibility that there is a difference in the effects of interests on behavior across sex (Harrop et al., 2018).

Lastly, due to the modest sample size, a small effect of diagnosis cannot be entirely excluded, especially for the exemplar task, even though our statistics reveal not even a trend in that direction. However, the sample size was adequate to demonstrate dependencies on category and array size, and a power analysis demonstrated that had there been a substantial effect of diagnosis, it would have been detected on at least one measure nearly 100% of the time in the category task, independent from power on the exemplar task. The power analysis used the actual sample sizes for each task, and a hypothetical effect size that was driven by the central question we posed: whether the abnormal interest pattern in ASD subjects could be viewed as merely a consequence of altered search (either a loss of efficient search for faces, or a replacement of efficient search for faces by efficient search for special interests). The reason that high power could be achieved with a relatively small subject pool is that there was relatively little variability of the performance measures within each group (i.e., the error bars in **Figures 2**, **3** are relatively small.) The exemplar task had much lower power than the category task for scenario (i), which makes sense given that performance differences between faces and houses were only found for the accuracy measure. For scenario (ii), while the power was lower and the direction of the faces-interests performance differential was reversed, a hypothetical effect of diagnosis was still detected 71% of the time.

CONCLUSION

In conclusion, individuals with ASD do not show large differences in early visual perception to intense interests compared to NT controls. The findings, while null, suggest that if there are abnormalities in the visual system in individuals with

ASD, they are not detectable at the level of visual search with faces or interest images. Further, despite enhanced day-to-day time spent engaging and looking at one's interest in ASD, there does not seem to be a direct impact of these interests on the early visual system. Together the findings provide insight into the growing body of work to understand the ASD symptoms relating to intense interests.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article can now be found on the project's OSF page, https://osf.io/rtpwx/ and in the **Supplementary Material** as **Data Sheet S1**.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Weill Cornell Medicine Institutional Review Board. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

RMJ and BMS contributed to the conception of the study. BMS contributed to data collection and data storage. BMS and JDV contributed to statistical analysis. All authors contributed to the design, read, revised, and approved the submitted manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2020.582074/full#supplementary-material

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Neurotype-Matching, but Not Being Autistic, Influences Self and Observer Ratings of Interpersonal Rapport

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The Double Empathy Problem suggests that communicative difficulties between autistic and non-autistic people are due to bi-directional differences in communicative style and a reciprocal lack of understanding. If true, there should be increased similarity in interaction style, resulting in higher rapport during interactions between pairs of the same neurotype. Here, we provide two empirical tests of rapport, with data revealing whether self- and observer- rated rapport varies depending on the match or mismatch in autism status within a pair. An additional opportunity afforded by these data is to examine the effect of the autism status of the rater on the perceived rapport between matched and mismatched pairs. In Study 1 72 participants were allocated to one of three dyad conditions: autistic pairs (n = 24), non-autistic pairs (n = 24) and mixed pairs (n = 12 autistic; n = 12 non-autistic). Each participant completed three semi-structured interactions with their partner, rating rapport after each interaction. Non-autistic pairs experienced higher self-rated rapport than mixed and autistic pairs, and autistic pairs experienced higher rapport than mixed pairs. In Study 2 (n = 80) autistic and non-autistic observers rated interactional rapport while watching videoed interactions between autistic pairs, non-autistic pairs, and mixed pairs (n = 18, a subset of participants in Study 1). Mixed pairs were rated significantly lower on rapport than autistic and non-autistic pairs, and autistic pairs were rated more highly for rapport than non-autistic pairs. Both autistic and non-autistic observers show similar patterns in how they rate the rapport of autistic, non-autistic, and mixed pairs. In summary, autistic people experience high interactional rapport when interacting with other autistic people, and this is also detected by external observers. Rather than autistic people experiencing low rapport in all contexts, their rapport ratings are influenced by a mismatch of diagnosis. These findings suggest that autistic people possess a distinct mode of social interaction style, rather than demonstrating social skills deficits. These data are considered in terms of their implications for psychological theories of autism, as well as practical impact on educational and clinical practice.

Keywords: autism, Double Empathy Theory, rapport, interaction, communication, neurodiversity

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INTRODUCTION

Rapport is defined by mutually experienced co-ordination, positivity, and attentiveness within a social interaction (Tickle-Degnen and Rosenthal, 1990). It is marked by a harmony and affinity between two people (Bernieri, 2014), and is a key component in constructing and developing successful interpersonal interactions (Cappella, 1990). As rapport relates to the quality of a relationship between two people, it is distinct from many other psychological constructs which are situated within individuals, rather than within interactions (Bernieri, 2014). Feelings of rapport can be influenced by social context with individuals from the same or similar social groups reporting higher rapport (Miles et al., 2011), even when those groups are defined by arbitrary or minimal criteria (Tajfel, 1981; Macrae and Bodenhausen, 2000; Brewer, 2007). Non-verbal and verbal communicative behaviors, including facial expressions, eye contact, postural mirroring, and tone play an important role in building rapport in people presumed to be neurotypical (Tickle-Degnen and Rosenthal, 1990); while not exhibiting these behaviors is related to poorer rapport (Richmond and McCroskey, 1995; Grahe and Bernieri, 1999; Hove and Risen, 2009). As difficulties with processing and expressing verbal and non-verbal social cues amongst autistic individuals have been well documented (Bottema-Beutel et al., 2019; Sasson et al., 2020), we might expect this to subsequently impact upon their development of rapport with others.

Autism is typically characterized by differences in social communication and interaction (American Psychiatric Association, 2013) compared with neurotypical norms. Popular attempts to explain autism, such as accounts like theory of mind (Frith, 2001), executive functioning (Ozonoff et al., 1991), or social motivation (Chevallier et al., 2012) adopt a deficit-based model. For instance, theory of mind explanations propose that social difficulties arise from a cognitive deficit residing in the autistic person preventing them from being able to infer, understand, or predict the behavior and intentions of others (Baron-Cohen et al., 1985; Gernsbacher and Yergeau, 2019). Experimental research showing that autistic people are unable to attribute mental states to others is believed to underlie autistic difficulties in social communication (Frith, 2001). Specifically, theory of mind deficits in autistic individuals has been linked to difficulties in identifying facial expressions (Uljarevic and Hamilton, 2013), and tone of voice (Rutherford et al., 2002). Additionally, autistic people have differences in frequency and patterns of eye contact, and postural and behavioral mirroring (Senju and Johnson, 2009; Hamilton and Marsh, 2013). Given these behaviors are thought to be related to rapport, it would be expected that interactions with and between autistic people would elicit low rapport. Applying a deficit model framework to paired interactions, autistic people should have the same difficulties interacting with autistic and non-autistic people (due to impairments in social communication) but difficulties would be compounded when two autistic people interact. A hypothesis based on this framework would predict that rapport between two non-autistic people would be highest, rapport between two autistic people would be lowest, and rapport between an autistic person and a non-autistic person would sit between these extremes.

Until recently, approaches to studying autism have been framed by neurotypical definitions of being social (Heasman and Gillespie, 2019a) and yet those with autism have a divergent neurotype, which often makes their mode of social communication different (Kapp et al., 2013). Increasingly, deficit-based paradigms are challenged by ideas grounded in the social model of disability, which proposes that autistic difficulties emerge as a result of systemic barriers in society (Kapp et al., 2013). There is increasing evidence suggesting that non-autistic people contribute to difficulties in interactions between autistic and non-autistic people (e.g., Edey et al., 2016; Sheppard et al., 2016; Sasson et al., 2017; Heasman and Gillespie, 2019a; Crompton et al., 2020a,b; Keating and Cook, 2020). This phenomenon has been conceptualized through the Double Empathy Problem, a theory which suggests that communicative difficulties between autistic and non-autistic people are due to bi-directional differences in communicative style and a reciprocal lack of understanding (Milton, 2012; Milton et al., 2018). The Double Empathy Problem contrasts with more traditional models of interaction in autism (Frith, 1994; Chevallier et al., 2012) and the diagnostic criteria (American Psychiatric Association, 2013; World Health Organization, 2020), which emphasize pervasive deficits in social interaction that are inherent in autistic populations. Instead, it suggests that difficulties arise due to a mismatch between autistic and non-autistic interaction styles, resulting in a decrement in social understanding on both sides.

Empirical support for the Double Empathy Problem is based on two strands of research. One area of research has explored non-autistic people's difficulties in interacting with autistic people. Non-autistic people are less accurate at deciphering the facial expression of autistic people (Sheppard et al., 2016) and struggle to interpret autistic people's mental states (Edey et al., 2016). Struggling to read autistic social cues is related to non-autistic people liking autistic people less (Alkhaldi et al., 2019), and non-autistic people are less willing to interact with autistic people (Sasson et al., 2017). Non-autistic people are also less likely to want to spend time or interact with autistic people than with non-autistic people (Morrison et al., 2020). These biases against autistic individuals are formed quickly by non-autistic people, and do not change with increased exposure (Sasson et al., 2017). Non-autistic people overestimate how egocentric autistic family members are (Heasman and Gillespie, 2018), while also overestimating the helpfulness of their own behaviors toward autistic people (Heasman and Gillespie, 2019b). Taken together, this body of research provides evidence that autistic social difficulties may be in part due to the perceptions of, and judgments made by, non-autistic people.

The second research focus has been to examine inter-autistic communication and interaction. There are distinctive features of interactions between autistic people (Heasman and Gillespie, 2019a; Granieri et al., 2020), and autistic people qualitatively report that their interactions with other autistic people are more comfortable and easier compared with interactions with non-autistic people (Crompton et al., 2020a). Though autistic people

may perceive other autistic people as being more awkward, less attractive, and less socially warm than non-autistic people, autistic people still express interest in future interactions with other autistic people (DeBrabander et al., 2019; Morrison et al., 2020), suggesting that autistic people base their social judgments on fundamentally different criteria to non-autistic people. Indeed, autistic people are less likely to find non-typical social behaviors in other autistic people problematic (Sng et al., 2020). Autistic people disclose more personal information to other autistic people, feel close to other autistic people (Morrison et al., 2020), empathize more with autistic people, and are more motivated to help them than non-autistic people (Komeda et al., 2019). While little is known about the mechanisms that underlie comfortable interactions between autistic people, autism-specific communication styles are associated with more positive first impressions by other autistic people (Granieri et al., 2020).

The Double Empathy Problem suggests that difficulties in interaction occur due to a lack of reciprocity between different neurotypes, and proposes that there will be increased reciprocity, and therefore higher rapport, between people of the same neurotype. According to the Double Empathy Problem, it would be hypothesized that rapport between autistic pairs and non-autistic pairs would be better than rapport within mixed autistic and non-autistic pairs.

Rapport has been measured using combinations of associated characteristics, such as warmth, empathy, understanding, friendliness and genuineness between those in the interaction (Tickle-Degnen and Rosenthal, 1990). Studies of rapport in dyadic interactions typically examine either self-rated questionnaires (i.e., each participant in the interaction rates the rapport they felt in their interaction, e.g., Frisby and Martin, 2010), or observer-rated questionnaires (i.e., observers watch video clips of dyads interacting, and rate the rapport between the two participants, e.g., Hall et al., 2009). While self-rated rapport can give an indication of one's personal experience of a social interaction, this judgment may be prone to biases (Pronin et al., 2004). Observer-ratings however may allow for a complementary, and more objective assessment of interpersonal interaction rapport between pairs of individuals.

In this paper we aim to contrast the deficit model framework with the Double Empathy Problem by conducting two studies assessing rapport between pairs of autistic adults, pairs of non-autistic adults, and mixed pairs where one person was autistic and one was non-autistic. Study 1 included self-rated rapport, as experienced during task-based dyadic interactions where each person's diagnosis status (autistic or non-autistic) was known by the other. Study 2 involved autistic and non-autistic observers rating rapport for videoed informal interactions between autistic pairs, non-autistic pairs, and mixed pairs. In this study the observers were blind to the diagnostic status of the participants engaging in social interaction within the videos. If social interaction difficulties experienced by autistic individuals were due to a mismatch in communication style, as posed by the Double Empathy Problem, we would expect the lowest ratings of rapport in mixed pairs in Studies 1 and 2. If however, rapport ratings were lowest in the autistic dyads (in both studies) these findings may align better with a deficit framework. A further component of both studies is the inclusion of autistic and non-autistic raters in each, which allowed us to explore whether rapport is judged similarly (both for self and others) within these two populations. If autistic individuals fail to pick up on appropriate social cues during or while viewing a social interaction, we would expect their judgments of rapport to differ from non-autistic individuals.

STUDY 1: SELF-RATED RAPPORT IN AUTISTIC, NON-AUTISTIC, AND MIXED PAIRS

Ethics and Recruitment

This study was carried out in accordance with the British Psychological Society's Code on Human Research Ethics. Experimental procedures for Study 1 were reviewed and approved by the University of Edinburgh Research Ethics Committee. All participants provided written informed consent prior to participating. Participants were recruited through community networks, social media, and local autism organizations.

Participants

Seventy-two adults participated: twenty-four adults in each of the autistic, non-autistic, and mixed groups. The mixed group therefore included 12 autistic and 12 non-autistic participants. A prospective power analysis was run, indicating 95% power to detect a medium effect of 0.5 at the standard 0.05 alpha error probability with a sample size of 66. The three groups were matched on age, gender, years of education, and IQ (**Table 1**). All spoke English to a native level and did not have a clinical diagnosis of social anxiety disorder. Participants also completed the Wechsler Abbreviated Scale of Intelligence II (WASI-II) (Wechsler, 2011), a measure of IQ, with all participants scoring within a typical range. Demographics are presented below based on dyad types for the purposes of the study, and demographic data based on the individual data (autistic, and non-autistic participants, $n = 36$ in each group) are shown in **Supplementary Material 1** for additional context.

Thirty-three autistic participants reported having received a diagnosis by a clinician. An additional three participants self-identified as autistic. Participants who self-identified as autistic also scored above 32 on the Autism Quotient (AQ) (Baron-Cohen et al., 2001) and above 72 on the Ritvo Autism-Aspergers Diagnostic Scale-Revised (Ritvo et al., 2011) indicating not only high levels of autistic traits but also a self-rating above a diagnostic threshold. All non-autistic participants scored below 32 on the AQ, indicating low levels of autistic traits (Baron-Cohen et al., 2001).

Materials and Procedure

All participants took part in three experimental tasks using a diffusion chain method (Crompton et al., 2020b). This procedure

TABLE 1 | Descriptive statistics and group comparisons [Mean (Standard Deviation)] for Study 1 participants on demographic variables, IQ, and autistic traits.

	Non-autistic (n = 24)	Autistic (n = 24)	Mixed (n = 24)	Comparisons
Age	37.92 (14.39)	37.33(13.13)	35.25 (10.76)	$X^2(2) = 0.27$, $p = 0.87$
Gender	21F, 3M	18F, 3M, 3NB[b]	18F, 6M	Fisher's exact test $p = 0.17$
Years of Education	17.83 (1.52)	17.44 (2.80)	17.12 (1.98)	$X^2(2) = 1.83$, $p = 0.40$
IQ – WASI-II[a]	115.04 (11.78)	114.42 (16.89)	117.79 (13.62)	$F(2,69) = 0.38$, $p = 0.68$
Autism Quotient	13.21 (5.44)	35.58 (6.18)	26.88 (14.27)	$X^2(2) = 32.26$, $p = 0.001$
Age of Diagnosis	NA	30.55 (12.72)	30.89 (10.20)	$X^2(1) = 0.36$, $p = 0.85$

[a]*Wechsler Abbreviate Scale of Intelligence -II.* [b]*Non-binary.*

involves a series of dyadic interactions in which an individual first observes a researcher complete a task, and then completed that task with a second participant. The second participant then completed the task with a third participant, and so on, until an eighth participant completes the task. In effect this allowed for 7 dyadic interactions between participants per chain (and thus yielding 63 interactions in total; 21 autistic, 21 non-autistic, and 21 mixed interactions). Only two participants were in the same room, and interacting, at any one time. Each chain of eight participants attended a different research day, hosted at the University of Edinburgh Division of Psychiatry.

Before the study commenced, participants were aware whether they were in an autistic, non-autistic, or mixed dyad. Participants did not meet before the first task started, and were isolated in separate rooms whilst they waited for their turn to take part in the study. The first dyadic task involved building a tower out of spaghetti and plasticine (Caldwell and Millen, 2008), the second involved sharing a fictional story (see Crompton et al., 2020b), and the third involved participants creating geometric animal shapes from a Rubiks Twist (TM). Each task took between 1 and 5 min, and participants interacted with each other freely while completing each task.

After each task, participants indicated their feelings of rapport using a 100-point scale with five dimensions: ease, enjoyment, success, friendliness, and awkwardness (reverse scored). Participants indicated a score for each dimension by drawing a cross on a horizontal line, indicating a scale from 1 to 100. The five dimensions had a Cronbach's alpha of 0.93, and so were summed to create a single scale of interactional rapport for use in subsequent analyses.

Design

This study used a between-groups design, comparing self-rated rapport in autistic, non-autistic, and mixed groups.

Results

For each dyad, a pair mean rapport score was calculated to reflect the overall rapport experienced by both participants in each dyadic interaction. This was calculated as the average of the rapport scores of both participants within each pair for each task. There was no significant interaction between the three dyadic tasks and the three groups (see **Supplementary Figure 1**), and so a summed mean was used in subsequent analyses, calculated as the mean of the pair's mean rapport scores for each of the three tasks (minimum = 0, maximum = 500).

The summed pair mean rapport scores met assumptions of normality and homogeneity of variance, and a subsequent one-way ANOVA found a significant difference in overall rapport between the three groups [$F(2,60) = 19.89$, $p < 0.001$. *Post hoc* comparisons using Tukey's HSD indicated that the non-autistic group experienced higher self-rated rapport than the mixed ($p < 0.000001$) and autistic group ($p < 0.05$), and the autistic group experience higher self-rated rapport than the mixed group ($p < 0.001$) see **Figure 1**].

Subsequent analysis explored potential effects of the participant's neurotype (autistic or non-autistic) and the social context (i.e., whether participants were in a matched chain with participants of the same neurotype, or a mixed chain with participants from a different neurotype) on self-rated rapport (**Figure 2**). A two-way ANOVA showed an effect of neurotype, with lower ratings of rapport in the autistic group [autistic mean = 370.38, non-autistic mean = 406.62, $F(1,68) = 12.32$, $p < 0.001$], and an effect of social context, with lower ratings in the mixed group [mixed mean = 351.30, matched mean = 407.1, $F(1,68) = 25.97$, $p < 0.001$]. However, there was

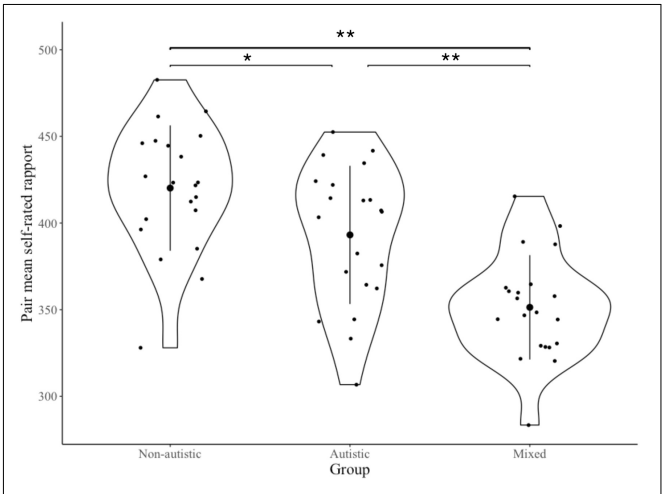

FIGURE 1 | Mean Pair self-rated rapport for non-autistic, autistic, and mixed groups. Bold dot indicates mean, line indicates standard deviation, and violin plot showing distribution of the data, *$p < 0.05$, **$p < 0.001$.

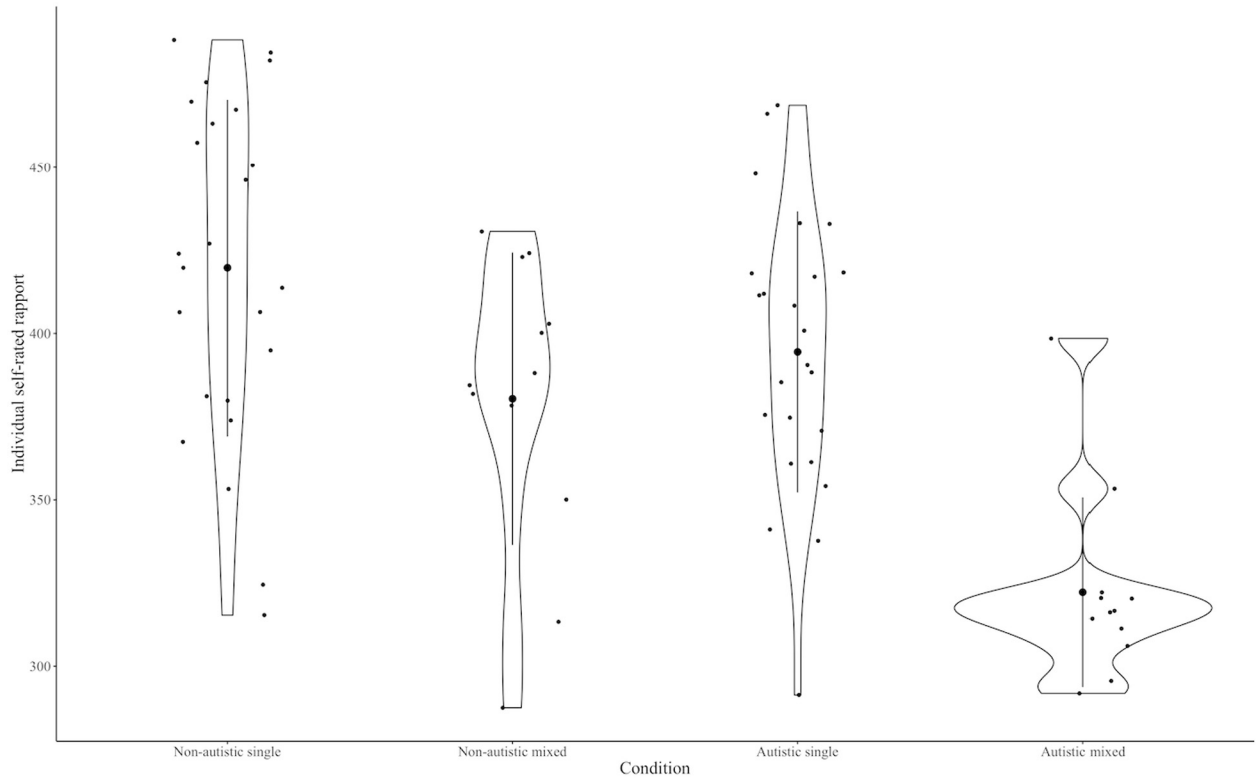

FIGURE 2 | Self-rated rapport for autistic and non-autistic participants in single and mixed social contexts. Bold dot indicates mean, line indicates standard deviation, and violin plot showing distribution of the data.

no significant interaction between rater neurotype and social context [$F(1,68) = 2.25$, $p = 0.13$].

Summary

This study examined how autistic and non-autistic people self-rated rapport with autistic and non-autistic partners. Participants completed short tasks with a partner, and afterward rated their experiences of rapport on a 5-dimensional scale.

Results showed that non-autistic pairs experienced higher self-rated rapport than autistic pairs, and both autistic and non-autistic pairs, and mixed pairs experienced lower rapport than both autistic pairs and non-autistic pairs. Regardless of individual neurotype, rapport is lower within mixed pairs compared with single neurotype pairs.

Additionally, examining the effect of the social context (i.e., whether in a matched or mixed-neurotype pair), showed that both autistic and non-autistic participants experienced lower rapport in mixed pairs. A lack of interaction with rater neurotype indicates that the lower rapport experienced in the mixed pairs is not driven by participants of a particular neurotype: both autistic and non-autistic participants had lower rapport within mixed pairs than in single neurotype pairs. However, given the small number of participants in each group when analyzing the data in this way ($n = 12$ each of autistic and non-autistic people in the mixed group), low statistical power may have contributed to the lack of a significant effect.

STUDY 2: OBSERVER RATED RAPPORT OF AUTISTIC, NON-AUTISTIC, AND MIXED PAIRS

Ethics and Recruitment

This study was carried out in accordance with the British Psychological Society's Code on Human Research Ethics. Experimental procedures were reviewed and approved by the University of Edinburgh Psychology Research Ethics Committee, the University of Nottingham (Psychology) Research Ethics Committee, and the University of Durham (Education) Research Committee. All participants provided written informed consent prior to participating. Participants were recruited through community networks, social media, and local autism organizations.

Participants

Study 2 included eighty participants (40 autistic and 40 non-autistic) recruited across three sites: 24 at the University of Edinburgh, 22 at the University of Durham, and 34 at the University of Nottingham. A prospective power analysis was run, indicating 95% power to detect a medium effect of 0.5 at the standard 0.05 alpha error probability with a sample size of 54. Two participants (one autistic and one non-autistic) were excluded after testing, due to having an AQ score which was

out of range (i.e., below or above 32 respectively) for their stated neurotype.

The final participant groups (39 autistic and 39 non-autistic individuals) were matched on age, gender and years of education. All spoke English to a native level. All non-autistic participants scored less than 32 on the Autism Quotient, indicating low levels of autistic traits (Baron-Cohen et al., 2001). Autistic participants were either clinically diagnosed ($n = 36$), or self-diagnosed ($n = 3$) and scored above 32 on the Autism Quotient (AQ) (Baron-Cohen et al., 2001). Demographic information for the autistic and non-autistic participants are shown in **Table 2**.

Materials and Procedure

The Paired Interaction Videos

Nine video stimuli were created for use in Study 2. These videos featured a subset of eighteen participants from Study 1. Videos featured three different pairs of autistic participants, three different pairs of non-autistic participants, and three different pairs of participants where one person was autistic and one was non-autistic (hereafter "mixed" pairs).

Each video featured a 2-min interaction between participant pairs (the first 2 min of a longer interaction, shortened to reduce task demand and length), who sat together at a table with their upper body and heads visible to viewers. Participants in the videos had been given a prompt sheet of paper providing basic statements to frame the interaction, for example "Tell me about where you live." Participants had not met each other before this interaction took place. After each interaction, participants in the videos completed the Rapport Measure, described in Study 1. Details about the demographics of video participants are outlined in **Supplementary Table 2**.

Observer Ratings of Rapport

Participants (observers) individually watched 3 videos, one from each dyad condition (i.e., autistic, non-autistic, mixed, with the order of presentation counterbalanced between observers). After each video, observers completed ratings of rapport using the same scale used in Study 1, measuring how easy, enjoyable, friendly, successful and awkward they thought the interaction between the observers in the video appeared, on a scale of 0–100. The observers did not know the diagnosis of individual people in the video, however they were aware that one or more people in the videos may have a diagnosis of autism. Observers watched each video start to finish before marking any responses to ensure they had fully seen and processed each interaction. Observers then completed the AQ (Baron-Cohen et al., 2001).

Design

This study used a mixed design, exploring how neurotype (autistic or non-autistic) affects observer-rated rapport of autistic, non-autistic, and pair dyads interacting in video clips. Researchers were blind to which pair was which in the videos, making it a double-blind study to minimize bias in the results.

Results

The five dimensions on the rating scale had a Cronbach's alpha of 0.91, and so were summed to create a single value of interactional rapport for use in subsequent analyses.

Initial review of the data revealed an outlier within the autistic group with lower overall rapport scores on the same neurotype pairings (autistic and non-autistic). A closer look at this individual's data showed no evidence of misunderstanding the scale (i.e., reversing coding) and as the results remained the same when conducted with the outlier removed it was decided to retain their data. Data in one of the dyad conditions (autistic pairs) were moderately skewed (−0.56) thus did not meet the assumption of normality. Another group (mixed pairs) did not meet the assumption of homogeneity of variances. However, as ANOVA is reported to be robust against small variations in the data distribution (Schmider et al., 2010) it was decided to proceed with parametric analysis.

A mixed 2 × 3 ANOVA was carried out to explore whether there were any group differences in how autistic and non-autistic participants judged rapport of social interactions between autistic, non-autistic, and mixed pairs (**Figure 3**). Results showed a main effect of dyad condition [$F = (1.67,127.57) = 24.07$, $p < 0.001$; non-autistic mean = 331.99, autistic mean = 364.25, mixed mean = 275.43]. Paired-sample *post hoc* tests revealed significantly lower rapport ratings for mixed pairs than autistic [$t(77) = -6.43$, $p < 0.001$] and non-autistic pairs [$t(77) = -3.81$, $p < 0.001$]. Furthermore, autistic pairs were found to have significantly higher ratings of rapport than non-autistic pairs [$t(77) = 3.38$, $p = 0.001$]. Between subject comparisons showed that both autistic and non-autistic observers did not differ in how they rated rapport in general across all videos [$F(1,76) = 0.428$, $p = 0.52$]. In addition, there was no significant interaction between rater diagnostic status and dyad condition [$F(2,127.57) = 0.75$, $p = 0.46$].

Though the small sample size prohibited formal comparison of self- and observer-rated rapport in Study 2, **Figure 4** illustrates how participants who created the video stimuli rated their

TABLE 2 | Descriptive statistics and group comparisons [Mean (Standard Deviation)] for Study 2 participants on demographic variables and autistic traits.

	Non-autistic ($n = 39$)	Autistic ($n = 39$)	Comparisons
Age	33.74 (13.31)	34.31(13.20)	$U = 1.28$, $p = 0.26$
Gender	25F, 14M	23F, 14M, 2NB[a]	Fisher's exact test $p = 0.56$
Years of Education	17.17 (2.26)	17.36 (3.12)	$U = 0.054$, $p = 0.817$
Autism Quotient	15.95(6.27)	37.50 (8.64)	$U = 48.43$, $p = 0.001$
Age of Diagnosis	NA	26.69 (12.77)	NA

[a]*Non-binary.*

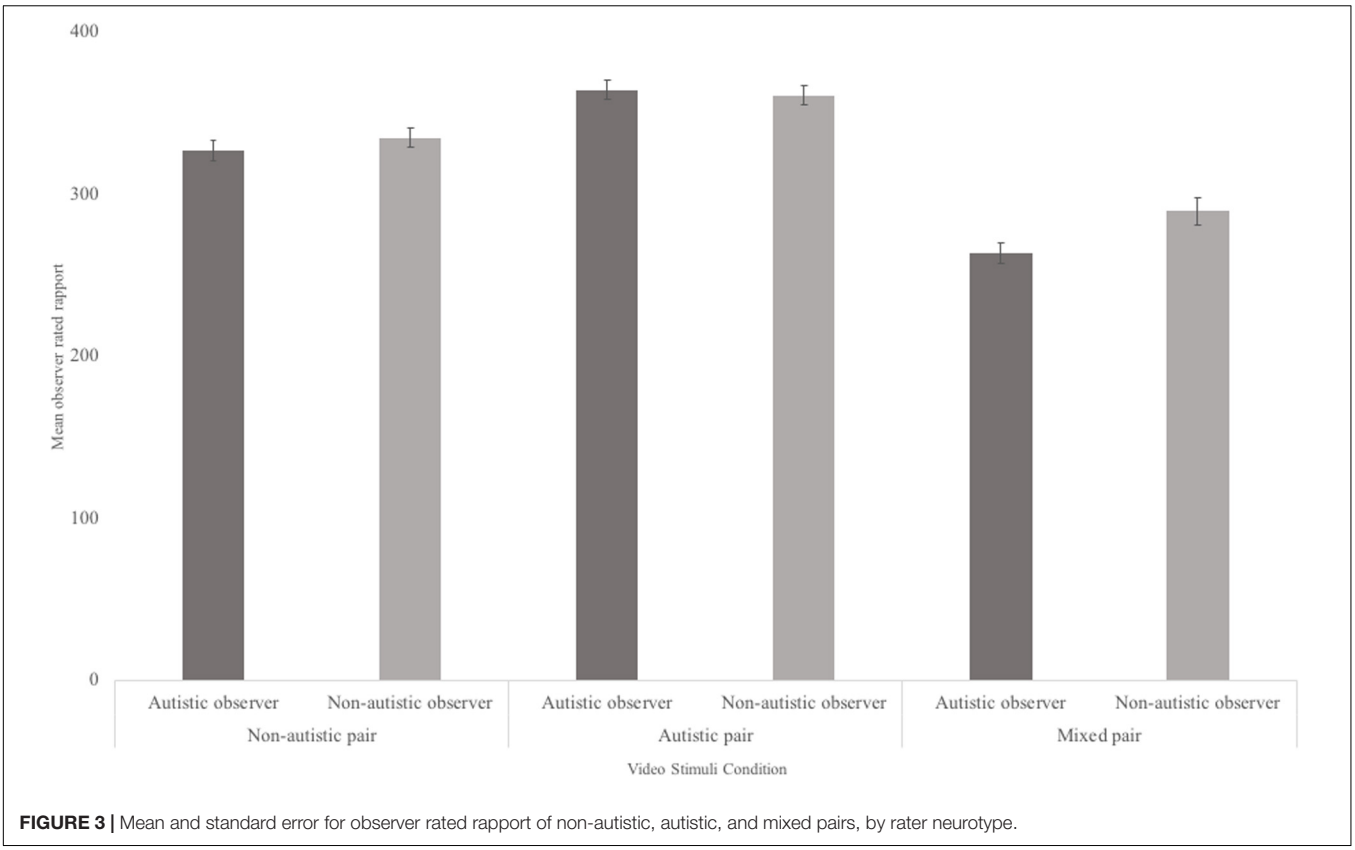

FIGURE 3 | Mean and standard error for observer rated rapport of non-autistic, autistic, and mixed pairs, by rater neurotype.

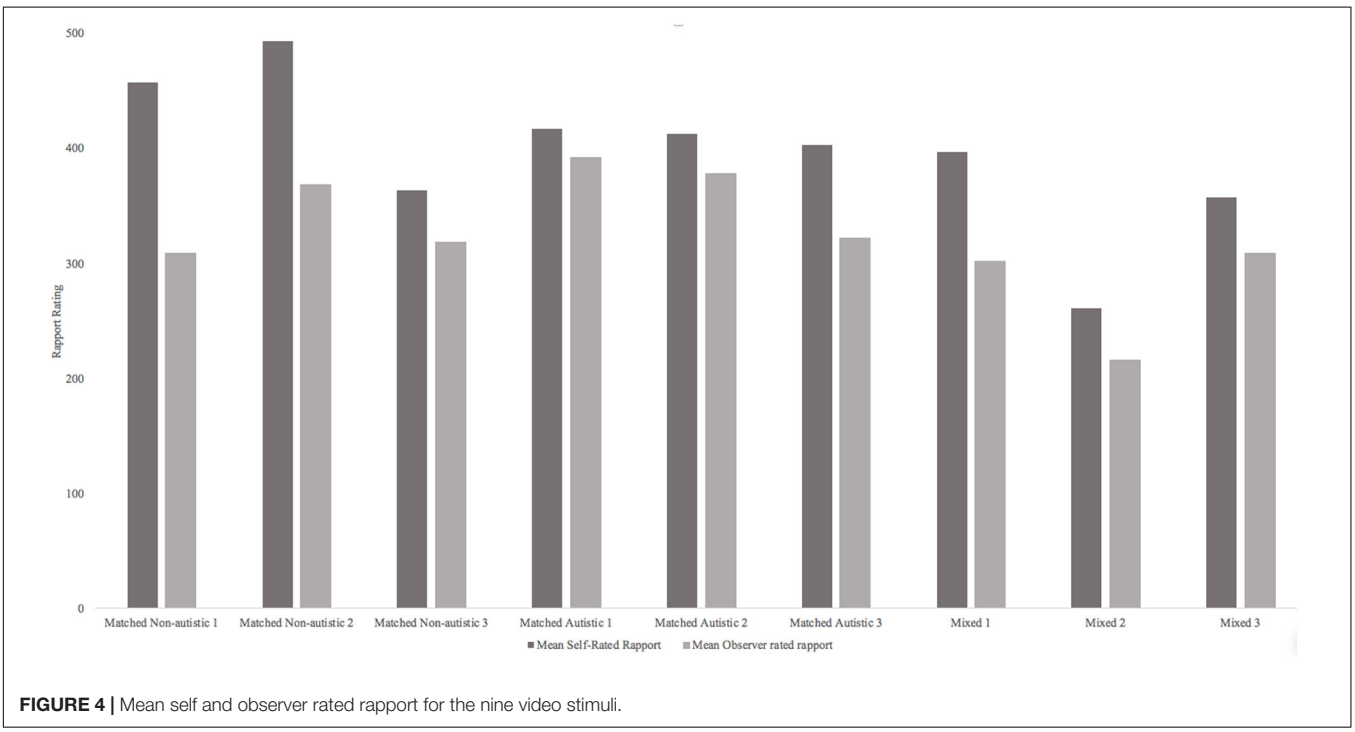

FIGURE 4 | Mean self and observer rated rapport for the nine video stimuli.

own rapport alongside how observer participants rated their rapport. Though these data are too limited for significance testing, it appears that autistic participants' self-ratings of rapport in matched autistic pairs are more similar to observer ratings of rapport (mean difference between self and observer rated rapport = 46.91). There is a greater difference between self-rated

and observer-rated rapport in matched non-autistic pairs (mean difference between self and observer rated rapport = 106.35).

Summary

Study 2 examined how observers rated rapport between autistic, non-autistic, and mixed pairs, and whether the diagnostic status of the rater (autistic or non-autistic) affected ratings of rapport in different neurotype pairs. The results indicate that participants, regardless of diagnostic status, give poorer ratings of rapport for mixed neurotype pairs than for matched neurotype pairs. This suggests a mismatch between neurotypes results in lower ratings of rapport, and that subtle verbal and non-verbal cues to rapport are similarly perceptible by autistic and non-autistic individuals. Interestingly, rapport scores were significantly higher for the autistic pairs than non-autistic pairs, indicating that the autistic dyads may display even greater social signals of shared enjoyment and ease when interacting with one another, as viewed by an external observer.

An exploratory comparison between participants' own judgments of rapport and an observer's ratings, suggests autistic participants' self-rating of rapport are more in line with others' ratings of rapport. There was a greater discrepancy between non-autistic participants' estimates of their rapport with a partner compared with observers' rating of the same social interaction.

DISCUSSION

Studies 1 and 2 examined perceptions of rapport between autistic pairs, non-autistic pairs, and mixed pairs. Though these two studies are not directly comparable as they involved different measures (self or other rated) of different social situations (task focused or informal chat) they both provide evidence that rapport between mixed pairs of individuals is significantly lower than in same neurotype pairs. These findings are consistent with our predictions and offer support for the Double Empathy Problem. A further common finding in both studies is that there were no differences in the pattern of rapport ratings between autistic raters and non-autistic raters. This suggests that autistic individuals discriminate between good and poor rapport between different dyad pairs like non-autistic pairs. In addition to these findings which are common to both studies, the results specific to each study and their implications will be discussed below.

In Study 1 it was demonstrated that self-rated rapport was poorer in mixed pair groups than in same pair (autistic-autistic; non-autistic-non-autistic) groups, as predicted according to the Double Empathy Problem. The results also showed that within the mixed dyad group both autistic and non-autistic people experience lower rapport when interacting with someone of a different neurotype. This provides evidence that the social difficulties autistic individuals experience when interacting with a non-autistic individual may at least partly be attributed to a mismatch in neurotype. Thus, social difficulties for autistic people may be relational in nature, rather than an individual impairment as posited by accounts which adopt a deficit model. These findings are in line with a recent review which argues that there is growing evidence to suggest that a theory of mind explanation for social difficulties in autism is questionable (Gernsbacher and Yergeau, 2019), and echoes findings from other research using a range of methodologies to examine the bi-directional nature of social interaction, considering communication as a joint experience rather than at the individual level (De Jaegher and Di Paolo, 2007; Bottema-Beutel, 2017; Sterponi and De Kirby, 2017). If rapport is constructed from subtle verbal and non-verbal cues during social interactions, then autistic individuals must be sufficiently able to detect these to discriminate between the mixed neurotype and same neurotype groups.

More broadly, these findings fit with the wider psychological literature on in-group/out-group effects (e.g., Tajfel, 1979). Social identity theory suggests that inter-group behaviors are based on perceived group status differences. Thus, if someone identifies as being part of the same group as someone else (in the case of this research – autistic people with other autistic people, or non-autistic with other non-autistic people) they may be more motivated to achieve positive results (i.e., high self-rated rapport) (Tajfel et al., 1979). In contrast, perceiving someone as being of a different group to you (in the case of this study where diagnostic status was known within mixed pairs), participants may be less motivated to have positive interactions and high self-rated rapport. Though the effect of neurotype group identity on social behavior has not been explored, when neurotypical children are assigned to different arbitrary groups (e.g., green team, blue team), they show reduced imitation of those in their outgroup, just as autistic children show reduced imitation of neurotypical children (van Schaik and Hunnius, 2016). This presents the possibility that reduced social engagement exhibited by some autistic people may be explained by a lack of identification with people from other groups (i.e., non-autistic people).

A further finding is that autistic pairs' self-rated rapport was significantly lower than non-autistic pairs self-rated rapport. There are several reasons why this may be the case. First, autistic pairs may experience lower rapport than non-autistic pairs due to differences in processing social information. Interpersonal interactions are a rich source of social information, and it is possible autistic individuals may be placing greater emphasis on some information more than others, or have their rapport limited by the volume of interactional processing going on (Murray et al., 2005). Second, due to well-documented autistic differences in social cognition (e.g., Sasson et al., 2020) autistic people may underestimate their rapport due to negative self-perception of their social skills (Hull et al., 2017) or lower self-perceived social competence (Jamison and Schuttler, 2015). Poor self-perception may also be the consequence of having a history of negative social interactions with individuals. Future research could ask autistic individuals to assess their overall level of social competence to see if this predicts self-rated rapport on a specific dyadic interaction. Third, autistic people could make rapport judgments on dimensions not assessed by the scale used in this study. Autistic people may have a distinctive way of interacting and building rapport with others (Heasman and Gillespie, 2019a), and may make social judgments using non-traditional criteria (Morrison et al., 2020), and thus their self-rated rapport may not be well assessed by the dimensions included in this scale. Finally, autistic people may be less impacted by social desirability

bias than non-autistic people (Kirchner et al., 2012), who may inflate their self-rated rapport scores to be viewed positively by the experimenter (Krumpal, 2013).

Interestingly, although Study 2 replicated the finding of reduced rapport in mixed neurotype pairs, it showed that observer-ratings of rapport indicated the opposite pattern to self-ratings in same neurotype pairs: autistic pairs were viewed as having higher interactional rapport than non-autistic pairs or mixed pairs, by both autistic and non-autistic observers. Whilst the finding of poorer rapport ratings in the mixed dyad groups as in Study 1 is again consistent with the Double Empathy account of autism, the finding of even higher ratings in the autistic pairs than non-autistic pairs is surprising. In this study, observers were blind to the neurotype of those in the videos although the participants themselves knew the diagnosis of the partner they were interacting with. One possible explanation for greater perceived rapport amongst autistic pairs could be that they immediately had something in common with the other individual (i.e., a diagnosis of autism) which may have helped them feel more at ease with one another from the start. Research showing individuals who have similar life experiences have greater social connection than those with different lived experiences supports this idea (Reagans, 2011). Although in Study 1 autistic pairs were also privy to their partners' diagnosis status, the lower rapport ratings in the autism pairs (in relation to non-autistic pairs) may have been due to higher self-ratings in the non-autistic group. Our exploratory analysis comparing self and other ratings of rapport (**Figure 4**) offers support for this interpretation.

As Study 2 involves observer ratings of rapport it is important to consider the findings in relation to the broader literature on person perception. Autistic people are perceived as being more awkward and less socially warm than non-autistic people (DeBrabander et al., 2019; Morrison et al., 2020), and being difficult to read is related to being perceived unfavorably by observers (Alkhaldi et al., 2019). In Study 2, rather than asking observers to rate the characteristics of individuals, observers rated the interpersonal rapport between two people sharing an interaction. Our findings contrast somewhat with previous findings of negative perceptions of autistic individuals, and it may be that interactions offer a different perspective. As observer ratings of rapport are enhanced by stable (compared to unstable) interpersonal coordination (Miles et al., 2009), it could be that pairs of the same neurotype have similar interpersonal styles, which translate into high rapport. Autistic interactions may follow a distinctive and unconventional pattern which function to effectively facilitate mutual understanding (Heasman and Gillespie, 2019a), and it is interesting that both autistic and non-autistic viewers rate autistic pairs as having high interactional rapport using our five dimensional measure. Future work may look to identify specific verbal and non-verbal markers of interactional rapport in autistic and non-autistic interactions. While the current study illustrates that there are differences in rapport, more detailed coding of interactions may begin to explore *why* rapport is better for autistic and non-autistic people. As approaches to studying autism are framed by non-autistic definitions of being social (Heasman and Gillespie, 2019a), and autistic people have a divergent neurotype, which often makes their mode of social communication different (Kapp et al., 2013), it is essential that any future coding schemes are co-designed with autistic people to be sensitive to and incorporate autistic social behaviors.

This study does have limitations, which could be addressed by future research in this area. First, as Studies 1 and 2 have some differences in design, we are restricted in the comparisons that we can draw between the two, and in how far we can contrast self-rated and observer-rated rapport. In Study 1, the interaction was more goal-oriented, whereas Study 2 was purely conversational. However, a similar pattern of findings across both studies does suggest a robust effect in different contexts which warrants future research. Second, though fully powered to detect moderate effects, the sample size was relatively modest, and only a small number of videos were used in Study 2. Future replications should use a range of videos representing a range of ages, genders and ethnicities to ensure that they are representative of the wider community.

Third, these studies did not use a standardized measure of rapport, as a measure that was appropriate to use for both self and observer rated rapport with adults who did not know each other could not be identified, and in addition, no rapport measures have been validated for autistic respondents. Our measure assessed core rapport domains identified in Tickle-Degnen and Rosenthal (1990) empirical and theoretical work on rapport, and creating a bespoke self-rating measure including these core domains is not atypical in rapport research (e.g., Bernieri et al., 1996; Lakin and Chartrand, 2003). However we cannot fully ensure the validity of the rapport measure used. If future work pursues this line of enquiry, a measure of rapport should be developed and validated for use with neurodiverse samples.

Fourth, participants in Study 1 and those who were filmed to create the stimuli videos for Study 2 were aware of the diagnostic status of the person with whom they were interacting, which could have affected their behavior and perceptions of rapport. As participants were aware of the diagnostic status of their partner, it is possible that both autistic and non-autistic people may have experienced higher rapport within single neurotype pairs because of perceived similarity or familiarity with their interlocutor. Autistic people may feel more comfortable with other autistic people (Crompton et al., 2020a), and non-autistic people may feel more comfortable with other non-autistic people (Cage and Burton, 2019; DeBrabander et al., 2019) and this may be enhanced by being aware of the diagnostic status of the other person in the interaction. Being aware of the diagnostic status of the person with whom they were interacting may have changed participants' behavior, however, previous research has shown that when non-autistic people know that they are interacting with an autistic person, they attempt to behave in a helpful way (Heasman and Gillespie, 2019b), and sharing diagnostic information results in greater acceptance of autistic people (Sasson and Morrison, 2019). As such, it may be hypothesized that there may be an even larger effect on rapport between mixed and single neurotype pairs if participants were blind to the diagnostic status of their partner. Although in some contexts diagnostic status may be known between individuals (e.g., peer-support groups, educational setting), at other times it may be unknown

(e.g., asking a shop assistant for help). Therefore, it will be important for future research to replicate the study with participants blind to the diagnostic status of their interaction partner.

Finally, the sample may not be representative of the wider autistic community, as all participants had an IQ within a normal range, and the sample had a large proportion of female participants. As autistic males are less likely to camouflage (Hull et al., 2020), this may impact rapport, though aligning with non-autistic expectations of what autism is may result in even lower rapport in the mixed pairs.

These findings suggest that autistic difficulties in building rapport are not a deficit within an autistic individual, and instead arise within interactions with non-autistic people. Further research exploring social difficulties within and between autistic and non-autistic people could have a significant impact on our theoretical and clinical understanding of autism based on a Double Empathy framework. Specifically, our findings challenge current diagnostic criteria and theoretical framing of autism. The finding that rapport is improved between autistic people strengthens calls for peer support for autistic people (Iemmi, 2017; Crane et al., 2020), particularly since a sense of belonging is a protective factor against suicide (Pelton and Cassidy, 2017). In an educational context, these findings challenge peer mediated support practices which specifically involve pairing autistic children with non-autistic peers who are meant to act as social "role models" (Chang and Locke, 2016). In light of the current findings one should reconsider the goal of this form of peer-mediated practice, and perhaps instead emphasize the mutual benefits of interpersonal interactions between mixed neurotypes in learning about diversity in communication. Future research is needed to identify and examine the specific behaviors that facilitate rapport between autistic people, which may in turn improve interactions between people of different neurotypes.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the University of Edinburgh Psychology Research Ethics Committee, the University of Nottingham (Psychology) Research Ethics Committee, and the University of Durham (Education) Research Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

CC: study design, data collection, data analysis, and led manuscript writing. MS and HA: data collection and revised manuscript. SF-W: project creative and scientific design, data analysis, and revised manuscript. EF: project creative and scientific design, revised manuscript. DR: project creative and scientific design, data analysis, and co-wrote the manuscript. All authors contributed to the article and approved the submitted version.

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ORIGINAL RESEARCH
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A Developmental Profile of Children With Autism Spectrum Disorder in China Using the Griffiths Mental Development Scales

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The purpose of this study was to profile the mental development of children aged 18 to 96 months with autism spectrum disorder (ASD) using the Chinese version of the Griffiths Mental Development Scales (GMDS), and to explore the relationships between developmental levels and ASD severity, the sex of the child and the age of ASD diagnosis. Children with ASD (n = 398; 337 boys, 61 girls) were recruited and ASD severity evaluated using the Autism Behavior Checklist and the Childhood Autism Rating Scale, while the GMDS was used to evaluate the children's mental development. Study participants were divided into groups according to GMDS general and subscale quotients, ASD severity, sex, and age. The majority of groups divided according to the GMDS quotients exhibited an unbalanced distribution in respect of the six domains of the GMDS and there were significant differences within the six subscale quotients. Autism severity, sex and age had significant effects on the overall level of development of autistic children. The quotients recorded for the children with more severe ASD were significantly lower than those for the children with less severe ASD. A markedly higher proportion of developmental delay was recorded for girls than boys in relation to the performance subscale. The locomotor quotient decreased in line with age at diagnosis, while autism severity and age had significant effects on the general and subscale quotients and sex had a significant effect on performance quotient. Children with ASD exhibit an uneven cognitive development profile, and their overall developmental levels are affected by autism severity, sex and age. Specific cognitive domains differ according to sex in children with ASD. Locomotor skills tend to decrease according to the age at diagnosis for autistic children aged 18 to 84 months. Autism severity and age are also associated with the level of functioning in different cognitive areas. These findings contribute to define the cognitive developmental profiles of children with ASD.

Keywords: autism spectrum disorder, children, developmental assessment, griffiths mental development scales, mental development

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Abbreviations: ASD, autism spectrum disorder; IDs, intellectual disabilities; ADOS, autism diagnostic observation schedule; GMDS, griffiths mental development scales; DSM-5, diagnostic and statistical manual of mental disorders; ABC, autism behavior checklist; CARS, childhood autism rating scale; GQ, general quotient; AQ, locomotor quotient; BQ, personal-social quotient; CQ, hearing and language quotient; DQ, eye-hand coordination quotient; EQ, performance quotient; FQ, practical reasoning quotient.

INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental condition, and individuals with the condition typically exhibit a range of atypical social interactions, communication difficulties, the presence of repetitive and stereotyped behavior, and restricted interests. The worldwide prevalence of the condition is thought to be between 1% and 3% of the general population, with a proportional distribution of four or five males to one female (American Psychiatric Association, 2013; Christensen et al., 2019). The sex imbalance in prevalence may be related to the underlying neurobiological mechanism. In addition, there may be sex differences in the autistic symptoms and cognitive development level of children with ASD, leading to the under-recognition and under-diagnosis of girls with ASD, exaggerating the sex imbalance (Van Wijngaarden-Cremers et al., 2014; Carpenter et al., 2019). Some community-based studies indicated that the true estimate of the ratio is likely to be closer to 3:1 (Honda et al., 2005; Kim et al., 2011; Idring et al., 2012).

The etiological factors remain largely unknown, but epigenetic factors, such as histone modification, DNA methylation and non-coding RNA, and the gut–microbiota–brain axis have been theorized to play an important role in ASD etiology (Martinez-Gonzalez and Andreo-Martinez, 2019; Andreo-Martinez et al., 2020; Yoon et al., 2020). Individuals with ASD often experience additional developmental disorders, with roughly 30% of those with ASD exhibiting other intellectual disabilities (IDs) (American Psychiatric Association, 2013). Children with more severe ASD (the nature and extent of ASD-related characteristics, henceforth referred to as the severity of ASD symptoms) generally have lower social adaptation abilities and require more support (Gardner et al., 2018). Additionally, autistic children with low developmental levels require more early intensive intervention to promote their developmental progress (Hinnebusch et al., 2017), and children with ASD who do not receive diagnoses of IDs and other developmental disorders may experience poorer developmental outcomes (Miller et al., 2019).

Children with ASD often exhibit unbalances with respect to their cognitive processing according to developmental assessment (Li et al., 2019). For example, compared with typically developing children, those with ASD can show significant difficulties concerning their relational and phonological working memory capacities (Ring et al., 2016; Habib et al., 2019). Their local visual information processing may be superior to that of typically developing children, but no differences between the two groups have been found with regard to global visuospatial performance (Muth et al., 2014; Nilsson Jobs et al., 2018). In addition, there may be sex differences in terms of the relative cognitive structures or developmental levels of individuals with ASD. Abilities corresponding to visual attention to detail in boys described as having high-functioning ASD were found to be superior to those of girls in Bölte et al.'s (2011) study, while Matheis et al. (2019) observed that girls aged 17–37 months who had ASD exhibited less communication difficulty but greater motor challenges compared to boys in the same age group. A further consideration is that the relative developmental levels of children with ASD may be related to the age at which they are diagnosed. For instance, Licari et al. (2020) analyzed the motor abilities of 2,084 autistic children younger than 6 years old, and found that approximately 30% of the children met the criteria for motor difficulties, and that the prevalence of motor difficulties increased with the age of diagnosis.

The Autism Diagnostic Interview–Revised (Rutter et al., 2003), the Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2000), and the Diagnostic Interview for Social and Communication Disorders (Wing et al., 2002) are the diagnostic tools that are most commonly used in relation to ASD (Randall et al., 2018). Children diagnosed with ASD may need to choose different types of educational centers, according to the severity of ASD symptoms and their cognitive levels. Therefore, before receiving intervention or education, they will require a standardized overall developmental assessment, with the purpose of refining the clinical diagnosis. More importantly, children's relative strengths and weaknesses can be identified at this stage, in order to facilitate the development of a constructive personalized intervention plan.

The Chinese versions of the Wechsler Primary and Preschool Scale of Intelligence (Wechsler, 2002) and Wechsler Intelligence Scale for Children (Wechsler, 2004) are commonly used intelligence tests in China, but they are only applicable to children aged between four and a half and 16 years old, and are not suitable for use in evaluating younger children (Gong and Dai, 1988; Yang, 2016). The Gesell Developmental Schedule, the Children's Neuropsychological and Behavior Scale, and the Griffiths Mental Development Scales (GMDS) are all diagnostic assessment tools commonly used in China to evaluate the development of children aged up to 6–8 years old. Of these, though, while the original version of the Gesell Developmental Schedule has been refined and updated, it has not been revised in the past 20 years for use in Chinese contexts (Yang, 2016), and the Children's Neuropsychological and Behavior Scale is a local assessment tool in China for which only the Chinese psychometric properties are currently available (Li et al., 2019).

Accordingly, the GMDS is the instrument that is generally used for evaluating developmental progress in children from birth up to 8 years of age (Luiz et al., 2001). It can be utilized for many clinical applications, such as predicting future developmental outcomes or evaluating the impact of epilepsy, antiepileptic drugs, and congenital heart disease surgery on infants' cognitive development (Dittrich et al., 2003; Randò et al., 2005; Bromley et al., 2010; Doyle et al., 2012). Moreover, the GMDS can be utilized as an intelligence test, in that the overall score ("general quotient") corresponds to an IQ score, and so it can be used to investigate the prevalence of ID in autistic children (Postorino et al., 2016; Scandurra et al., 2019).

Age at diagnosis, degree of atypicality, and level of intelligence may be key factors in predicting long-term developmental outcomes for individuals with ASD (Coplan and Jawad, 2005). Consequently, it is important to assess developmental levels in preschool and in early childhood. Prior studies have been primarily concerned with the cognitive characteristics of school-age children and adults with ASD, and the Wechsler (2002, 2004) intelligence scales have been most often used in such research (Hidding et al., 2015; Kanai et al., 2017; Kim and Song, 2020).

Although some previous studies have used the GMDS as an intelligence tool with which to assess the prevalence of ID in autistic children (Postorino et al., 2016; Scandurra et al., 2019), few have reported the motor, language, social, and reasoning abilities of children with ASD through reference to the GMDS subscale, or how these levels of ability correlate with the autism severity, age, and sex of the assessed individual.

The Chinese version of the GMDS was revised for use with Chinese children in 2016, based on the 2006 update to the 1996 version of the GMDS (Luiz et al., 2006). A cross-cultural comparison study confirmed that GMDS was well adapted to a Chinese context and could reliably be used to assess development in Chinese children from birth to 8 years old (Tso et al., 2018). Li et al. (2020) found that the scale had good reliability and validity in the evaluation of children aged 3 to 8 years old with ASD. Recently, He et al. (2019) used the scale to assess the mental development of children with ASD in China–specifically, to analyze the correlation between the developmental levels and eye movement characteristics of 21 preschoolers with ASD. To date, there is currently a dearth of literature characterizing the cognitive motor and social profiles of autistic children in China. The purpose of the present study is to profile the developmental levels of 398 children (18 to 96 months old) with ASD across the different areas of the GMDS, in order to explore whether there are unbalances between these areas, and to analyze the correlations between the developmental levels measured and the severity of the ASD, the sex of the assessed child, and the age at which they were first diagnosed with ASD as an attempt to provide a theoretical basis for interventions and educational decision-making in respect of children with ASD.

MATERIALS AND METHODS

Participants

The study's participants were recruited from a group of children who had exhibited signs of ASD and were being evaluated for the first time at the Child Developmental and Behavioral Division of the First Hospital of Jilin University in Changchun, China, during the period March 2018 to December 2019. Initially, all of the children with suspected ASD were examined through reviews of their current health, developmental history, and family history, as well as through a clinical physical examination and parental interviews carried out by at least two developmental pediatricians with reference to the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5) criteria (American Psychiatric Association, 2013). In addition, an ADOS administration was undertaken by trained developmental pediatricians. Ultimately, 398 children from this group were found to fulfill the DSM-5 criteria for ASD and had positive results from their ADOS administration.

All participants in the study completed assessments designed to evaluate both their developmental levels and the severity of their ASD symptoms. The children had a mean age of 41.6 ± 15.6 months (range: 18–96 months), and the overall study cohort was composed of 337 boys and 61 girls with a 5.5:1 male-to-female ratio. All children were examined for common comorbidities such as epilepsy, and, following comprehensive medical observation and neuroimaging, genetic metabolism, chromosome, and other related examinations, we excluded children with Rett syndrome, fragile X syndrome, genetic metabolic disorders, and other neurological conditions such as epilepsy.

In the past, we have published outcomes from analyses of 114 boys and 25 girls enrolled in this study (Li et al., 2019). Prior to participation, all of the legal guardians of the children with ASD had given written informed consent. The Ethics Committee of the First Hospital of Jilin university approved this study (No: 2017-314).

Measurements

Evaluation of ASD Symptoms

The severity of ASD symptoms in the children participating in this study was assessed by a developmental pediatrician using the Autism Behavior Checklist (ABC) (Krug, 1980) and the Childhood Autism Rating Scale (CARS) (Schopler et al., 1980). The ABC and the CARS are both commonly used assessment scales in China in clinical practice and ASD research.

The ABC is an unstructured behavior questionnaire that is completed by the child's parent or caregiver. The checklist features 57 items, covering five aspects of atypical behavior: sensory, relating, body concept and object use, language, and self-care. The score for each item ranges from 1 to 4, and total scores for the ABC range from 0 to 158, with higher scores indicating increased levels of ASD symptoms. A typically developing child's ABC score should be less than 47 (Krug, 1980; Krug et al., 1993). The Chinese version of the ABC has been found to have good psychometric properties (Yang et al., 1993), and with a cut-off score of 50 of the checklist, autism was screened from the normal population with a sensitivity of 0.97 and a specificity of 1 (Yang et al., 1993).

The CARS is a 15-item observational scale. Each item was graded by the developmental pediatrician on the basis of the symptom criteria, with a rating of 1 denoting "*normal*," 2 "*mild*," 3 "*moderate*," and 4 "*severe*." Typically developing children exhibit CARS scores of lower than 30 (Schopler et al., 1980), and higher scores indicate more severe ASD symptoms. The reliability coefficient (Cronbach's alpha) was 0.94, and the correlation coefficient between the scale scores and clinicians' ratings was 0.80, indicating good reliability and validity for the CARS (Schopler et al., 1980).

ASD Diagnostic Evaluation

In order to further corroborate the diagnosis of ASD, all children with suspected ASD underwent an ADOS assessment, performed by a developmental pediatrician who had received training and qualified in ADOS evaluation. In this study, the Chinese version of the ADOS was used, which was revised based on the second edition of ADOS (ADOS-2; Lord et al., 2012). The ADOS is a play-based, semi-structured assessment tool for assessing current autistic behaviors. It consists of four different modules, which are selected by the child undergoing assessment according to their current expressive language level. Each module has a specific diagnostic algorithm for two domains: social affect and restricted and repetitive behavior. Overall, an ADOS evaluation

takes about 45 min to complete. The total score of each module has a cut-off point corresponding to whether it conforms to the diagnosis of ASD.

Assessment of Mental Development

The GMDS was used to evaluate the developmental levels of the autistic children. Three experienced developmental pediatricians who had been formally trained in the test and were qualified to use it for research evaluation participated in this study. The Chinese version of the GMDS was revised based on the 2006 version of the GMDS and featured normative data relating to China (Luiz et al., 2006; Tso et al., 2018).

The GMDS measures a child's abilities through reference to the following six subscales: subscale A is the "locomotor" scale, measuring movement with regard to graded coordination, economy of effort, and postural control; subscale B measures "personal–social" abilities, covering growing self-awareness, independence, and social interaction; subscale C assesses "hearing and language," rating the child's ability to hear, listen, and comprehend, as well as to express themselves; subscale D appraises "eye and hand coordination," or visual competence with fine motor precision functionality; subscale E covers "performance" as it pertains to visual perception awareness, including working speed and precision; and subscale F corresponds to "practical reasoning," or a 2 to 8-year-old child's ability to use past learning experiences to solve problems, as well as their understanding of basic mathematical concepts and moral issues.

The mean of the general quotient (GQ) and each of the six subscale quotients is 100 points (SD = 15 points). The subscale quotients are calculated using the developmental age corresponding to each subscale divided by the actual chronological age and multiplying by 100. The GQ raw score is the sum of the subscales raw scores. A GQ or a subscale quotient $<$70 points ($>$2SD below the mean) is considered to indicate a significant delay in development, while a quotient $>$70 points indicates a mild or no delay (Cirelli et al., 2015). The Cronbach's alpha of the full scale of the Chinese version of the GMDS was 0.98, indicating a strong correlation between the subscales, while the subscales' Cronbach's alphas were all above 0.7, suggesting acceptable internal consistency (Tso et al., 2018).

Procedures

During the first visit to the hospital, children who had exhibited signs of ASD will receive an initial assessment of approximately 20 min by an outpatient developmental behavioral pediatrician, including current health, developmental history, and family history. For children suspected with ASD, the outpatient pediatrician would schedule an evaluation checklist containing ABC, CRAS, GMDS, and ADOS. On the day of the first visit, the parents of the participating children completed the ABC after being given instructions on how to do so by a developmental pediatrician in the evaluation room of the Child Developmental and Behavioral Division. At the same time, the pediatrician completed the CARS by observing the child's behavior and conducting an interview with their parent or guardian. If the child was in a good condition, a trained and qualified developmental pediatrician would complete the GMDS on the day of the first visit too. The GMDS assessment was performed in a quiet examination room or a training room approximately 20 square meters in size and with no distracting objects in the room during the course of the evaluation. A full GMDS evaluation takes approximately one and a half hours to complete. If a child had an obvious emotional reaction during the evaluation, a new appointment could be made, but the evaluation had to be completed within 1 week. The ADOS was also usually completed within 1 week, with the certified developmental pediatrician completing it in an assessment room of approximately 20 square meters in size, and a full ADOS assessment took approximately 45 min for each child.

Statistical Analyses

The data were analyzed using SPSS Statistics, version 22.0 (IBM Corp., NY, United States). The normality of the data was analyzed using the Kolmogorov–Smirnov test. Continuous data were means $\pm$ SDs or P50(P25, P75) (i.e., median, 25th percentile, and 75th percentile measures), whereas categorical data were given as frequencies with percentages. Based on the GQ and five subscale quotients (i.e., those for the GMDS subscales B to F; subscale A was excluded for the weak correlation with cognitive structure), the study's sample of autistic children was subdivided into two groups, as follows. Children who received a GQ or a subscale quotient $>$70 points were assigned to a *higher developmental-level* group, while children who scored $<$70 points were allocated to a *lower developmental-level* group. Subjects in the former group were observed to have demonstrated mild or no delay in their general development or in one of the domains measured by the GMDS, whereas children in the latter group had exhibited a significant delay in terms of their general development or in respect of one of the domains measured by the GMDS. The Kruskal–Wallis H test was used to compare the differences of quotients in various fields of the GMDS within each group. Mixed ANOVA was used to compare the mean differences within the six subscales (GQ and subscale A to E), as well as differences in overall level of development among factors such as autism severity, sex and age.

According to their total CARS scores, children with ASD were assigned into groups as follows. Children with a total CARS score of fewer than 32 points were considered to have *less severe* ASD (n = 226, mean CARS score = 28), whereas ASD was considered to be *more severe* in children whose scores were equal to or higher than 32 points (n = 172, mean CARS score = 35). A 32-point cutoff was chosen as it was the mean CARS score for all of children with ASD in the study's sample. An independent samples t-test and a non-parametric Mann–Whitney U test were used to compare the continuous data of the two groups. Normally and non-normally distributed data were analyzed using Pearson's and Spearman's correlation coefficient tests, respectively.

An independent samples t-test, a non-parametric Mann–Whitney U test, and chi-squared tests were used for comparing variables between the different sex subgroups. Cohen's d was calculated between the variables to represent the magnitude of the differences. The Kruskal–Wallis H test was also used to compare

the differences of the GQ and subscale quotients of the GMDS among the six age groups.

Multi-way ANOVA was used to compare the effects of autism severity, sex and age on the GQ and six subscale quotients of the GMDS. All tests were two-sided, with $P < 0.05$ as the significance threshold.

RESULTS

Autism Severity and Developmental Levels of the Participants

The total ABC score of the participants was 51.7 ± 16.9, and the total CARS score was 31.5 ± 4.4. The subscales and GQ scores of the GMDS of the participants are summarized in **Table 1**. Developmental delays were considered to be present when a GQ or subscale quotient was at least 2 *SD* below the mean (a GQ or a subscale quotient <70). Data for children exhibiting developmental delays in the different domains of this scale are presented in **Table 1**. As these children had limited language ability and developmental progression, measurable practical reasoning scores (i.e., obtained using the GMDS subscale F) were only available for 99 children (25%); no scores in this domain were provided for the other 299 children (75%).

The Developmental Profiles of the Participants in the Two Developmental Level Categories

Figure 1 plots the developmental profiles of the study's sample of autistic children in the two developmental level categories, grouped by GQ or subscale quotients. As can be seen in **Figures 1A,B,D,E**, participants in both the higher and the lower developmental-level groups (grouped by GQ, BQ, DQ, and EQ, respectively) demonstrated an unbalanced distribution of GMDS results in six areas. The relatively best results were found in relation to the locomotor and performance subscales (i.e., A and E, respectively) while the lowest was in respect of the hearing and language subscale (C). However, as illustrated in **Figures 1C,F**, the developmental quotient distribution curves of the higher developmental-level groups (grouped by CQ and FQ, respectively) were relatively flat. There were no significant differences observed among the six subscale quotients in the higher developmental-level group of **Figure 1C** ($n = 66$, $H = 4.862$, $P = 0.433$), indicating that there was no developmental imbalance found for the children in this group. In the higher developmental-level group of **Figure 1F**, except for BQ (mean = 77.2, *SD* = 14.7), no significant differences were found among the remaining subscale quotients ($n = 43$, $H = 2.420$, $P = 0.659$).

TABLE 1 | GMDS assessment results for children diagnosed with ASD by the age of 8 (96 months)[a].

Quotient (subscale letter label)	Mean ± *SD*	Delay[b] *n* (%)
General (GQ)	62.2 ± 17.2	279 (70.1%)
Locomotor (AQ)	75.7 ± 17.9	146 (36.7%)
Personal–social (BQ)	57.4 ± 19.3	297 (74.6%)
Hearing and language (CQ)	48.0 ± 23.0	332 (83.4%)
Eye–hand coordination (DQ)	63.3 ± 19.0	251 (63.1%)
Performance (EQ)	66.7 ± 23.4	227 (57.0%)
Practical reasoning (FQ)[c]	70.7 ± 23.4	56/99 (56.6%)
Delayed in two or more domains	–	316 (79.4%)

[a] *N = 398.*
[b] *A GQ or a subscale quotient <70.*
[c] *FQ was measured for 99 children (2–8 year olds) in the sample.*

Differences Within Subscales of GMDS and the Effects of Autism Severity, Sex and Age on Overall Developmental Level

A 2(autism severity) × 2(sex) × 7 (chronological age group) × 6(subscales) ANOVA gave a significant difference within the six subscale quotients of the GMDS ($F = 43.191$, $P < 0.001$, $\eta^2 = 0.359$), indicating an unbalanced distribution of GMDS results in six domains of the autistic children. The test also gave significant major effects of autism severity ($F = 6.819$, $P < 0.001$, $\eta^2 = 0.081$), sex ($F = 3.188$, $P = 0.008$, $\eta^2 = 0.040$) and age ($F = 12.252$, $P < 0.001$, $\eta^2 = 0.159$) on the overall level of development of autistic children. Age and autism severity ($F = 2.138$, $P = 0.048$, $\eta^2 = 0.033$) had interacting effects on the overall level of development; however, sex and age ($F = 2.040$, $P = 0.072$, $\eta^2 = 0.027$), sex and autism severity ($F = 0.175$, $P = 0.972$, $\eta^2 = 0.002$) had no interacting effects.

Developmental-Level Differences With Less Severe Versus More Severe ASD

The developmental levels of children with ASD of different levels of severity are detailed in **Table 2**. The ages at diagnosis of those in the more severe group were significantly lower than those in the less severe group. No significant differences in sex between the two groups. Total ABC scores and total CARS scores of children in the more severe group were significantly higher than those of the less severe group. The GQ and six mean or median subscale quotients (AQ–FQ) recorded for children with more severe ASD levels were significantly lower than those for children with less severe ASD levels.

Table 3 presents the correlation coefficients of the total ABC and CARS scores alongside age at diagnosis and the GMDS GQ and subscale quotients. Total ABC and CARS scores were found to be negatively correlated with age at diagnosis, GQ, and the subscale quotients.

Sex Differences for Autism Severity and Developmental Levels

Relative levels of autism severity and the developmental quotients of children with ASD for both sexes are compared in **Table 4**. No significant differences were found in relation to age at diagnosis or total ABC and CARS scores between boys and girls in the study's sample. However, the GQ of boys was significantly higher than that of girls. Regarding the six subscale quotients, the DQ, EQ, and FQ of boys were found to be significantly higher than

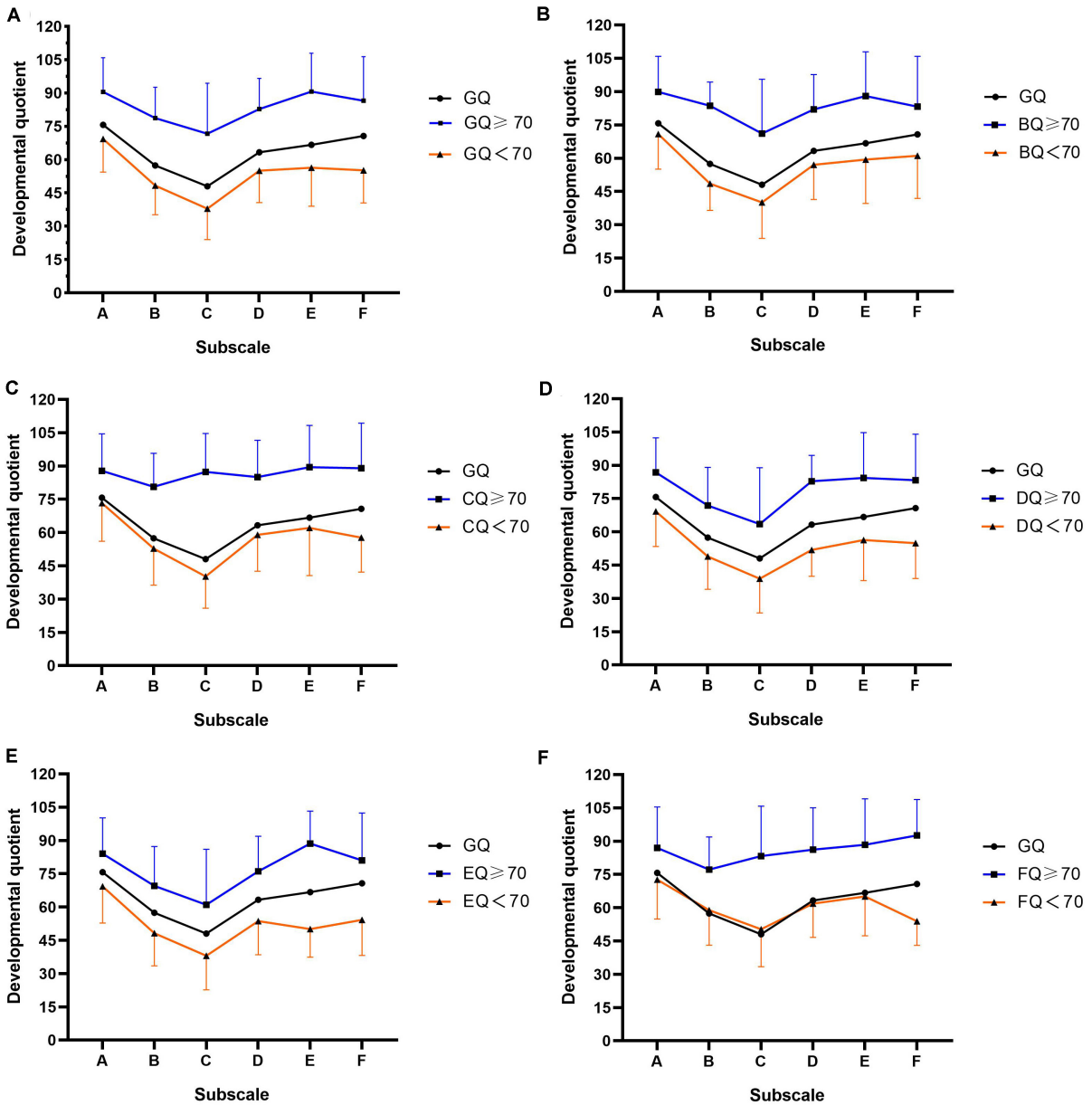

FIGURE 1 | Developmental profiles of autistic children included in this study plotted according to the two categorized developmental levels and grouped by GQ or subscale quotients. **(A)** Mean GMDS subscale GQ scores for the whole group and for each developmental level. Mean GQ score and mean developmental quotients on the subscales in the two levels grouped by: **(B)** GMDS subscale B (personal–social, BQ), **(C)** GMDS subscale C (hearing and language, CQ), **(D)** GMDS subscale D (eye–hand coordination, DQ), **(E)** GMDS subscale E (performance, EQ), **(F)** GMDS subscale F (practical reasoning, FQ). Standard deviations of the mean are represented in the figure by the error bars attached to each line of the two developmental levels.

those of girls, but no significant differences in AQ, BQ, or CQ were recorded between the two sexes.

The proportions of children of both sexes exhibiting delays in the different domains of the GMDS are summarized in **Table 5**. The proportions of children with developmental delays recorded in relation to the general, locomotor, personal–social, hearing and language, eye–hand coordination, and practical reasoning subscales did not differ significantly between boys and girls. However, the proportion of boys who were observed to have a developmental delay with reference to the performance subscale (EQ < 70) was significantly lower than that of girls.

Distribution of the Subscale Quotients of GMDS in Different Age Groups

We also analyzed variations in the developmental quotients of the GMDS in relation to the children's different age groups. There were only eight children (3 boys, 5 girls) in the sample aged between 84 and 96 months (i.e., 7 and 8 years old), and only

TABLE 2 | Developmental levels in children with less severe versus more severe ASD.

Variable	Less severe[a]	More severe[b]	t (Z)/χ^2	P
Age at diagnosis (months)	43.8 ± 15.6	38.7 ± 15.3*	3.28	0.001
Sex Boys Girls	198(87.6%) 28(12.4%)	139(80.8%) 33(19.2%)	3.477	0.062
Total ABC score	43.5 ± 14.6	62.5 ± 13.1*	13.4	<0.001
Total CARS score	28.4 ± 2.9	35.4 ± 2.2*	26.13	<0.001
General (GQ)	68.6 ± 16.5	53.7 ± 14.4*	9.38	<0.001
Locomotor (AQ)	79 ± 17.9	71.3 ± 16.9*	4.38	<0.001
Personal–social (BQ)	65.1 ± 17.6	47.2 ± 16.5*	10.33	<0.001
Hearing and language (CQ)	56.4 ± 23.5	36.9 ± 16.8*	9.23	<0.001
Eye–hand coordination (DQ)	69.6 ± 18.1	55.1 ± 17.0*	8.15	<0.001
Performance (EQ)	73.3 ± 22.3	57.9 ± 22.0*	6.86	<0.001
Practical reasoning (FQ)[c]	68 (55, 92)	57 (37, 76)*	(2.17)	0.03

[a] *n = 226.*
[b] *n = 172.*
[c] *FQ was measured for 99 children (2–8 year olds) in the sample: less severe, 83; more severe, 16.*
** Significantly different from values obtained for the less severe ASD group, $P < 0.05$.*

TABLE 3 | Correlation of total ABC and CARS scores with age at diagnosis and GMDS general and subscale quotients.

Variable	Total ABC score r_s/(r)	Total CARS score r_s
Age at diagnosis (months)	−0.19**	−0.21**
General (GQ)	(−0.38)**	−0.54**
Locomotor (AQ)	(−0.21)**	−0.26**
Personal−social (BQ)	(−0.45)**	−0.58**
Hearing and language (CQ)	(−0.34)**	−0.56**
Eye−hand coordination (DQ)	(−0.34)**	−0.48**
Performance (EQ)	(−0.27)**	−0.40**
Practical reasoning (FQ)	−0.21*	−0.41**

Spearman's rank correlation coefficient was used to obtain r_s; the Pearson correlation coefficient was used to obtain r.
**The correlation was significant, $P < 0.05$.*
***The correlation was significant, $P < 0.01$.*

TABLE 4 | Sex differences in autism severity and developmental levels of children with ASD[a].

Item	Boys[b]	Girls[c]	t (Z)	Cohen's d	P
Age at diagnosis (months)	41.5 ± 14.7	42.4 ± 19.9	0.326	0.05	0.746
Total ABC score	51.6 ± 16.7	52.6 ± 18.1	0.422	0.06	0.673
Total CARS score	31.3 ± 4.3	32.1 ± 4.7	1.011	0.18	0.312
General (GQ)	63.1 ± 16.7*	57.2 ± 19.5	2.440	0.33	0.015
Locomotor (AQ)	76 ± 17.3	73.8 ± 20.8	0.893	0.12	0.372
Personal–social (BQ)	58.1 ± 18.5	53.6 ± 22.8	1.649	0.22	0.100
Hearing and language (CQ)	48.4 ± 23.1	45.7 ± 22.7	0.855	0.12	0.393
Eye–hand coordination (DQ)	64.4 ± 18.3*	57.3 ± 21.9	2.704	0.35	0.007
Performance (EQ)	68.5 ± 23*	56.7 ± 23.4	3.656	0.51	<0.001
Practical reasoning (FQ)[d]	68 (55, 89)*	52 (37, 63)	(2.228)	–	0.026

[a] *P50(P25, P75), mean ± SD.*
[b] *n = 337.*
[c] *n = 61.*
[d] *FQ was measured for 99 children (2–8 year olds) in the sample: 91 boys and 8 girls.*
**Significantly different from values obtained for the girls, $P < 0.05$.*

TABLE 5 | Sex differences in the numbers of children exhibiting developmental delays based on GMDS quotients.

Subscale of GMDS (quotient letter label)	Boys[a] n (%)	Girls[b] n (%)	χ^2 n (%)	P n (%)
General (GQ)	234 (69%)	45 (73%)	0.463	0.496
Locomotor (AQ)	125 (37%)	21 (34%)	0.158	0.691
Personal–social (BQ)	251 (74%)	46 (75%)	0.024	0.878
Hearing and language (CQ)	278 (82%)	54 (89%)	1.359	0.244
Eye–hand coordination (DQ)	208 (62%)	43 (70%)	1.706	0.192
Performance (EQ)	181 (53.7%)*	46 (75%)	9.926	0.002
Practical reasoning (FQ)[c]	49/91 (54%)	7/8 (88%)	3.390	0.067

[a] *n = 337.*
[b] *n = 61.*
[c] *FQ was measured for 99 children (2–8 year olds) in the sample: 91 boys and 8 girls.*
**Significantly different from values obtained for the girls, $P < 0.05$.*

99 children were measured in relation to the practical reasoning domain (subscale F, which only applied to children between 2 and 8 years old). Accordingly, in order to avoid data bias caused by small sample sizes, these two groups were excluded from the data.

Table 6 shows the GQ and subscale quotients of the GMDS across six age groups. There were no statistically significant differences found regarding the mean scores of the GQ, BQ, and EQ between the six age groups. However, the mean scores of the AQ, CQ, and DQ were statistically significant between the different age groups. In addition, as can be seen in **Figure 2**, the mean scores of the AQ decreased with age at diagnosis, and there was a significant negative correlation between AQ and age at diagnosis (**Figure 2**) ($r = -0.310$, $P < 0.001$); however, there was no decreasing trend corresponding to age at diagnosis in the mean CQ or the DQ mean scores. With reference to the hearing and language subscale, the highest mean scores of CQ were for the 36–48 months group, and the lowest were for the 18–24 months group. In respect of the eye–hand coordination subscale, the highest scores for the DQ were in the 18–24 months group, while the mean scores of the remaining age groups fluctuated within the range of 58.3 to 64.3 points.

The Effects of Autism Severity, Sex and Age on the GMDS General and Subscale Quotients

The effects of autism severity, sex and age on the GQ and six subscale quotients of the GMDS using multi-way ANOVA

TABLE 6 | GMDS general and subscale quotients of in different age groups (months)[a].

Quotient	18–24[b]	24–36[c]	36–48[d]	48–60[e]	60–72[f]	72–84[g]	*H*	*P*
GQ	67.1 ± 13.4	62.6 ± 16.5	65.3 ± 18.1	59.5 ± 15.3	58.9 ± 19.0	59.7 ± 19.6	9.483	0.091
AQ*	87.5 ± 14.8	78.3 ± 16.2	75.8 ± 18.6	73.4 ± 15.9	67.4 ± 15.7	62.2 ± 16.3	41.227	<0.001
BQ	62.4 ± 18.1	56.7 ± 19.8	60.3 ± 19.7	57.1 ± 19.6	54.0 ± 14.2	52.5 ± 14.1	6.037	0.303
CQ*	41.9 ± 12.5	45.3 ± 23.5	55.7 ± 26.2	48.3 ± 17.5	49.5 ± 23.3	53.4 ± 24.9	17.632	0.003
DQ*	72.7 ± 16.0	64.3 ± 16.8	64.1 ± 19.5	58.3 ± 17.7	62.2 ± 25.6	64.1 ± 22.7	15.133	0.01
EQ	70.9 ± 23.2	68.1 ± 23.5	70.4 ± 23.2	62.4 ± 20.4	61.3 ± 23.7	64.2 ± 23.6	7.186	0.207

[a]*Mean ± SD.*
[b]*n = 34.*
[c]*n = 163.*
[d]*n = 92.*
[e]*n = 60.*
[f]*n = 27.*
[g]*n = 14.*
**Significantly different among the six age groups, P < 0.05.*

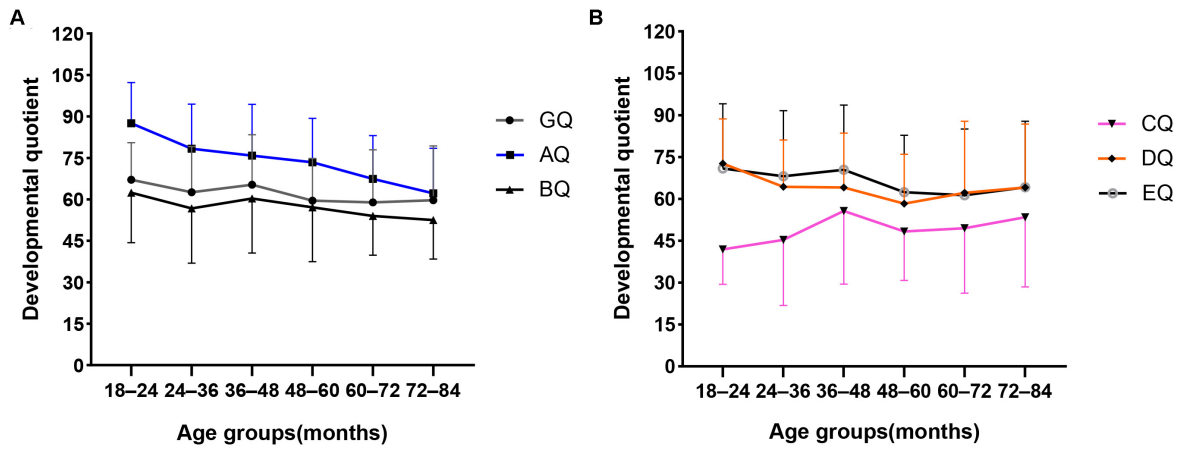

FIGURE 2 | Mean scores distribution of the GQ and A–E subscale quotients in different age groups. **(A)** Mean scores distribution of the GQ, AQ, and BQ in different age groups. **(B)** Mean scores distribution of the CQ, DQ, and EQ in different age groups. Standard deviations of the mean are represented in the figure by the error bars attached to each line.

test were detailed in **Table 7**. Autism severity had significant major effects on GQ and subscale quotients except FQ. Age had significant major effects on the general and six subscale quotients of GMDS. Sex had a significant major effect on EQ. No interacting effects were found among factors of autism severity, sex and age.

DISCUSSION

This study set out to profile the mental development of children with ASD between the ages of 18 and 96 months old. The relationships between developmental level and autism severity, sex, and age at diagnosis were also explored. Nearly 80% of the children included in this study were found to have comorbid developmental delays concerning two or more domains of the GMDS, a finding that is consistent with prior studies that have discovered that the majority of individuals with ASD have mild to moderate IDs, along with language difficulty (Postorino et al., 2016; Narzisi et al., 2018). In addition, similar to other studies, we found that most of the children exhibited a cognitive profile that typically encompassed uneven cognitive development, with relative strengths with regard to the locomotor and performance domains and weaknesses in respect of the hearing and language domain (Sandberg et al., 1993; Joseph et al., 2002); however, in the higher developmental-level group in this study, results pertaining to the hearing and language subscale (CQ > 70) and the six domains of the GMDS were relatively balanced, indicating that language difficulty was probably the main reason for the characteristically unbalanced cognitive profile.

In the higher level-development group's results for the practical reasoning subscale (FQ > 70), there was also no obvious unbalance in respect to the six GMDS fields–a finding that may be related to fact that there was found to be mild or no language difficulty in the children of this group, because the practical reasoning subscale incorporates mathematical concepts as well as ethics and moral issues requiring higher language comprehension ability. Clinically, children in these two groups are more likely to be described as having high-functioning ASD, with an average or above-average developmental quotient and no significant ID or language difficulty (Ousley and Cermak, 2014).

TABLE 7 | Effects of autism severity, sex and age on the GMDS general and subscale quotients.

Variable	GQ	AQ	BQ	CQ	DQ	EQ	FQ[a]
Autism severity	$F = 103.854$ $P<0.001$	$F = 36.379$ $P<0.001$	$F = 118.856$ $P<0.001$	$F = 75.983$ $P<0.001$	$F = 83.906$ $P<0.001$	$F = 53.642$ $P<0.001$	$F = 2.94$ $P = 0.09$
Sex	$F = 1.238$ $P = 0.267$	$F = 0.011$ $P = 0.915$	$F = 0.390$ $P = 0.533$	$F = 0.574$ $P = 0.449$	$F = 2.952$ $P = 0.087$	$F = 6.984$ $P = 0.009$	$F = 1.300$ $P = 0.257$
Age (group)	$F = 7.832$ $P<0.001$	$F = 15.121$ $P<0.001$	$F = 4.546$ $P<0.001$	$F = 3.094$ $P = 0.006$	$F = 8.415$ $P<0.001$	$F = 5.015$ $P<0.001$	$F = 4.053$ $P = 0.002$
Sex* Autism severity	$F = 0.476$ $P = 0.505$	$F = 1.384$ $P = 0.264$	$F = 0.078$ $P = 0.788$	$F = 0.450$ $P = 0.509$	$F = 0.309$ $P = 0.588$	$F = 0.313$ $P = 0.582$	–
Sex*Age	$F = 0.605$ $P = 0.703$	$F = 1.711$ $P = 0.285$	$F = 0.141$ $P = 0.975$	$F = 1.214$ $P = 0.418$	$F = 1.502$ $P = 0.333$	$F = 0.162$ $P = 0.966$	–
Age*Autism severity	$F = 1.911$ $P = 0.233$	$F = 0.746$ $P = 0.636$	$F = 1.475$ $P = 0.336$	$F = 3.603$ $P = 0.064$	$F = 2.461$ $P = 0.153$	$F = 1.882$ $P = 0.226$	–

Independent variables: Autism severity (less severe and more severe group); Sex (boys and girls); Age (seven age groups).
[a]*In some age groups, there was only one girl with FQ, so the interacting effects on FQ cannot be calculated.*

Mixed ANOVA analysis indicated a significant difference within the six subscale quotients of the GMDS, further verifying that the cognitive structure of autistic children was not balanced, and is simultaneously affected by the severity of autism, sex and age. Furthermore, age and autism severity had interacting effects on the overall level of development. These findings suggest that, before receiving intervention or education, autistic children need to undergo a standardized developmental assessment to identify their relative strengths and weaknesses, and to facilitate the choice of educational center and the formulation of a personalized intervention plan.

The GQ and six subscale quotients recorded for the group of children with more severe autism severity were significantly lower than those recorded for the group with less severe levels of ASD. Across the whole group, the GQ and six subscale quotients were negatively correlated with autism severity, suggesting that developmental level is closely correlated with symptom severity in autistic children. Among these, the personal–social and language domains showed a higher correlation with ASD severity, reflecting the close association between these two domains and the core symptoms of ASD. Early developmental levels, particularly the developmental quotient pertaining to the performance subscale, could predict later childhood IQ levels (Sutcliffe et al., 2010), and the results of our study would appear to further verify the hypothesis that autism severity increases with decreases of IQ (Mayes and Calhoun, 2011). These findings also suggest that ASD and its common comorbidity ID may overlap in pathogenesis (Coll-Tané et al., 2019). A lower IQ and the more severe social–communicative features of ASD are associated with lower adaptive functioning in the future (Tillmann et al., 2019), and so it is essential that interventions are developed to improve adaptive skills across different developmental levels and ASD severity. In addition, our study found that the age at diagnosis, for children with more severe ASD, was significantly lower than that of children in the less severe group, and the age was negatively correlated with ASD severity in the whole group. This reflects a common clinical phenomenon that children with more severe ASD symptoms often come to the hospital earlier for evaluation and diagnosis than children with less ASD severity.

In terms of prevalence, ASD has been established to be one of the neurodevelopmental disorders that is different according to sex (Mahendiran et al., 2019), a finding that is also reflected in this study, with a boy-to-girl ratio of 5.5:1. Although there was no significant sex difference in terms of age at diagnosis or autism severity, girls were recorded as having significantly lower scores in the GQ, eye–hand coordination, performance, and practical reasoning GMDS subscales than boys, and the proportion of girls with significant developmental delays in the performance subscale was higher, indicating sex differences in the developmental levels of autistic children. Moreover, boys with ASD may have better visuospatial skills than girls with ASD, since the performance subscale mainly measures visual perception abilities. Bölte et al. (2011) studied sex differences in relation to cognitive domains in 35 males and 21 females described as having higher-functioning ASD, and found that visual attention to detail in males with ASD was superior to that for girls, and proposed that this might be a potential basis for specific cognitive strengths in males with ASD, such as scientific or technical skills. Matheis et al. (2019) assessed the developmental functioning of 1,317 children with ASD aged 17–37 months through reference to the Battelle Developmental Inventory, and their results showed that females with ASD had greater motor difficulties and less communication challenges compared to males. The present study found that, although eye–hand coordination (fine motor) difficulties were more severe in girls, there were no sex differences in gross motor skills, personal social skills, or language skills. However, Duvall et al. (2020) research concluded that there were no sex differences concerning the cognitive abilities of young children with ASD aged 18–68 months. These different results may relate to the differences in sample sizes, sample ages, and test tools, which need to be further explored.

The distribution of the subscale quotients of the GMDS across different age groups suggested that locomotor skills tend to decrease in line with age at diagnosis. This pattern is consistent with findings from previous studies that motor difficulties become more pronounced with age (Landa and Garrett-Mayer, 2006; Lloyd et al., 2013; Licari et al., 2020). The transition from infancy to preschool child requires the acquisition of increasingly complex movement skills through increases in muscle strength, coordination, and stability (Licari et al., 2020). If a child has challenges in acquiring simple movement skills, it will be more difficult to acquire complex movement skills during subsequent

development stages. This may account for the relatively poorer locomotor quotients corresponding with an increasing age in the present study. Some prospective follow-up studies of high-risk infants across early development have found that motor difficulties in the infancy period is associated with later ASD diagnosis or ASD symptoms (Estes et al., 2015; Paquet et al., 2016; West, 2019). Therefore, motor difficulties may be an early marker preceding a diagnosis of ASD, but longitudinal follow-up studies are needed for further verification.

Although language skills and eye–hand coordination varied among the age groups in our study, they did not decline with age. Interestingly, in the language domain, the group comprising children aged 18–24 months had the lowest CQ scores, and the 36–48 months group had the highest CQ scores, with no significant fluctuation after 48 months. This could be explained by the fact that most children with ASD do not have meaningful language skills until 24 months old, and 24–48 months is a rapid period of language development in children with ASD. In turn, this might indicate that the level of language development at 48 months may predict the language prognosis in ASD. Brignell et al. (2018) found that language ability at 4 years and IQ rather than social communication skills influence the language prognosis in children with ASD. However, a longitudinal follow-up study is needed to verify this deduction. In respect of the eye–hand coordination subscale, the 18–24 months group recorded the highest DQ scores, with no significant fluctuation after 24 months. It may be that the test items in this domain before the age of 24 months are mainly based on perceptual observation, which does not require a high level of language comprehension. However, after the age of 2 years old, the need for language comprehension in this area is increased. Hence, the DQ scores of the later age group were lower than those of the 18–24 months age group.

In-depth exploration of the dataset using multi-way ANOVA demonstrated that autism severity and age had major effects on almost all GMDS subscales and GQ, indicating that children with ASD at different ages and with different levels of autism severity had different developmental levels in various areas of cognitive structure. In addition, sex had a major effect on performance quotient, further indicating that boys with ASD may perform better than girls in this domain, which mainly measures visual perception ability. To date, this is the first report in China on the effects of autism severity, sex, and age on the different cognitive structure domains measured using the GMDS. The present study contributes to describing the cognitive developmental profiles of children with ASD.

This study has several limitations, including the use of a cross-sectional research design. Although the developmental levels of ASD for different age groups were compared in the present research, the data obtained cannot be taken to represent the development trends of the same groups according to age. In addition, the developmental level of children with ASD at different ages was also affected by the context, drug or rehabilitation therapies and education. Therefore, longitudinal follow-up studies are needed to further verify the effect of age on the developmental level. For the comparison of sex differences in relation to developmental level, no typically developing children were included as a control group, and the ratio of boys to girls in this study was 5.5 to 1. Although some cognitive differences between boys and girls were detected, they are likely to be affected by this sex unbalance, which is another limitation of the study; however, given the identified sex and age differences in the relative development levels of autistic children, determining the most effective means with which to make up for these deficiencies, as well as how and when to select and provide the most appropriate interventions, will be an important future extension of our research in the future.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of the First Hospital of Jilin University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

H-HL and F-YJ participated in the design and definition of this study. C-XW, J-YF, and C-LL provided assistance for data acquisition and literature search. H-HL and C-XW performed the statistical analysis and drafted the manuscript. BW and J-YF carried out the manuscript editing. All authors have read and approved the content of the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2020.570923/full#supplementary-material

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A Brief Report: Community Supportiveness May Facilitate Participation of Children With Autism Spectrum Disorder in Their Community and Reduce Feelings of Isolation in Their Caregivers

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Children with autism spectrum disorder (ASD) participate at lower rates in their community, and their caregivers experience higher levels of stress, in comparison to families of typically developing (TD) children. The social model of disability positions the environment as the central issue when children with disabilities are unable to participate, yet little is known about the relationship between poor community support, reduced community participation in children with ASD, and caregiver stress. This study examined caregiver perceptions of community supportiveness for the community participation of 48 children with ASD (aged 5–12 years), alongside caregiver-reported child ASD symptom severity, adaptive functioning, and caregiver stress. Community supportiveness predicted child involvement, but not attendance, when child characteristics were held constant. Caregiver perceptions of low community supportiveness significantly predicted caregiver feelings of isolation. The importance of modifying community programs to better support inclusion of children with ASD is discussed.

Keywords: autism spectral disorder, community participation, children, caregivers, stress

INTRODUCTION

Children with autism spectrum disorder (ASD) often miss out on participating in activities such as community events, organized physical activity, informal interactions with other children, overnight visits or trips, and dance classes (Egilson et al., 2017; May et al., 2019), and their caregivers experience higher levels of stress than caregivers of typically developing (TD) peers (Hayes and Watson, 2013; Keenan et al., 2016). This reduced participation is a concern given community participation has been identified as a universal right for all children (UN General Assembly, 2007) and an important component of a child's social, physical, and psychological development (Eime et al., 2013; Howells et al., 2019, 2020; May et al., 2019).

Children with ASD experience many barriers to community participation as a function of their everyday challenges with social interactions and communication, under or over reaction to sensory input, a strong desire for predictability and routine, and repetitive patterns of behavior (American Psychiatric Association, 2013). A range of studies have identified links between increased ASD symptom severity and low community participation (Bar-Shalita et al., 2008; Hochhauser and Engel-Yeger, 2010; Shattuck et al., 2011; Thompson and Emira, 2011; Krieger et al., 2018; May et al., 2018). These include isolation or peripheral involvement due to challenges with peer social interactions (Shattuck et al., 2011; Krieger et al., 2018), and decreased attendance due to sensory impairments (Bar-Shalita, et al., 2008; Hochhauser and Engel-Yeger, 2010; Krieger et al., 2018; May et al., 2018), anxiety (May et al., 2018), and repetitive and restricted behaviors and interests (Thompson and Emira, 2011; May et al., 2018). Adaptive functioning describes personal and social skills that support an individual's ability to perform day-to-day activities independently (Sparrow et al., 2016) and encompass communication skills, daily living skills (linked to decreased community participation; Poon, 2011), socialization skills, motor skills (linked to decreased participation in community sports and leisure activities; Obrusnikova and Cavalier, 2011; May et al., 2018), and maladaptive behaviors (i.e., internalizing or externalizing behaviors) in reference to the ways in which greater difficulties with these skills disrupt day-to-day independence (Sparrow et al., 2016).

It has been argued from the perspective of the social model of disability that the environment plays a central role in determining whether children with disabilities and developmental challenges can participate fully (Shakespeare and Watson, 2002). As opposed to the medical model of disability, in which people are considered disabled as a function of their impairments, the social model shifts this focus to disability as a function of the barriers that prevents all people from being able to fully participate (Oliver, 2013). Consequently, proponents of this model are more interested in determining the ways in which environmental factors may inhibit or support participation than in identifying child "deficits" that may contribute to this inhibition. In keeping with this premise, research has explored community factors that inhibit the participation of children with ASD in their community. For example, adolescents with ASD who perceive the environment as low in safety and predictability have been found less likely to participate in their community (Krieger et al., 2018), and this is likely more pronounced in adolescents with ASD who have a higher need for adherence to routines and sameness (a symptom of ASD; American Psychiatric Association, 2013). Further, youths with ASD have reported that unclear implicit social demands act as a deterrent to their participation in community settings, as they feel intimidated when they are unsure how to understand, interpret, or react to social demands (Krieger et al., 2018), highlighting the likelihood that greater impairment in social skills will be linked to poorer participation in this population. Children with ASD can experience varying degrees of cognitive and communication challenges, and caregivers have reported that participation can be inhibited by the cognitive demands of community activities (Egilson et al., 2017). A number of other external factors that relate to both ASD symptom severity and adaptive functioning have been identified as barriers to the participation of children with ASD in their community, including lowered availability and suitability of appropriately trained community services and staff (Krieger et al., 2018).

Most previous research has not distinguished between child community participation measured as "attendance" and child community participation measured as "involvement." Attendance captures a child's presence within a program, but does not measure the depth of the participation, that is, how engaged and included they are (Imms et al., 2016). This is an important distinction, particularly when considering that ASD is characterized by challenges with initiating and responding to social interactions (American Psychiatric Association, 2013), in that they may attend regularly, yet not demonstrate the same level of involvement as their TD peers. Further, while research has identified ways in which symptom severity and adaptive behavior appear associated with their participation in community activities, there is an absence of research that examines both child characteristics and caregiver perceptions of community supportiveness in relation to both child attendance and involvement within a single sample. Examining both of these constructs together is important, as it is possible that mere attendance of a program may not improve social, physical, and psychological outcomes if involvement in the program is low. Further, understanding how the community environment supports or inhibits child participation is important for moving beyond a child deficit viewpoint to the social model of disability, as many of these factors may be amenable to modification. Participation in community activities such as organized and unstructured physical activity declines further in children with ASD as they move into adolescence (Simpson et al., 2019), highlighting the importance of early intervention in these settings to promote continued access to future participation opportunities. Community programs, such as organized physical activity, have been identified as promising psychosocial interventions for children with ASD (Rinehart et al., 2018) and identifying community barriers to participation may help inform modifications to community programs to support the full inclusion and participation of children with ASD, thereby improving social, physical, and psychological outcomes for children with ASD.

Understanding the mechanisms that may contribute to the relationship between caregiver stress and community participation is particularly important when taking into consideration the high rates of stress caregivers of children with ASD experience in comparison to caregivers of TD children (Hayes and Watson, 2013; Keenan et al., 2016). Caregivers of children with ASD tend to have fewer opportunities to engage in social interactions (Lecavalier et al., 2006; Myers et al., 2009) and face challenges in accessing community-based social supports (Sanders and Morgan, 1997). Further, caregivers who perceive social support received by themselves or their child as inadequate are more likely to experience high levels of stress (Gray and Holden, 1992; Sanders and Morgan, 1997; Siklos and Kerns, 2006; Siman-Tov and Kaniel, 2011). Indeed, research has found that a lack of social support, including stigmatization of a child's behaviors or characteristics, can result in caregivers of children with ASD withdrawing from social situations (Sanders and Morgan, 1997; Eaton et al., 2016), thereby experiencing increased stress

(Sanders and Morgan, 1997). Similarly, qualitative research has highlighted a lack of community support for the inclusion of children with disability as being one of the key challenges to caregiver well-being, with caregivers reporting experiencing high levels of stress and isolation, including feeling "labeled" by other parents, due to having to play the perpetual role of advocate for their child's inclusion across multiple settings (Resch et al., 2010). Previous research found that caregivers rate community social organizations as offering the least helpful support (Hall and Graff, 2011), however, while a large body of research has examined the relationship between caregiver stress and social support from family and friends, to our knowledge, there is no quantitative literature that specifically examines the relationship between community supportiveness and stress in caregivers of children with ASD. In the closest study identified, child ASD symptom severity and community supportiveness could account for 16% of the variance in family coping scores (Hall, 2012).

To our knowledge, however, no previous research has examined whether lower levels of community supportiveness is linked to reduced community participation in children with ASD while holding ASD symptom severity and adaptive behaviors constant. Further, little research has examined these relationships in relation to the dual constructs of participation – attendance and involvement. Similarly, while previous qualitative research has identified possible links between lower levels of community supportiveness and higher levels of caregiver stress, this has not been evaluated while controlling for variability in ASD symptom severity and adaptive behaviors.

The aims of the current study are two-fold:

1. To examine the relationship between caregiver perceptions of community supportiveness and child participation (attendance and involvement);
2. To examine the relationship between caregiver perceptions of community supportiveness and caregiver stress.

Based on research findings children or youth with ASD, and their caregivers, identify a range of external barriers (i.e., cognitive and social demands of activities) to community participation (Egilson et al., 2017; Krieger et al., 2018), it is predicted that higher supportiveness of the community environment will predict higher levels of child community participation and involvement when holding child characteristics constant. Further, based on research identifying links between social support and caregiver stress (Gray and Holden, 1992; Sanders and Morgan, 1997; Siklos and Kerns, 2006; Siman-Tov and Kaniel, 2011) and community support and family coping (Hall, 2012), it is hypothesized that reduced community supportiveness and the higher symptom severity and adaptive behaviors that are often associated with community participation will be predictive of higher levels of caregiver stress.

MATERIALS AND METHODS

Participants

The participants were 56 children aged 5–12 years and diagnosed with ASD, who were recruited as part of a larger pilot study examining the outcomes of participation in a community football program in metropolitan and regional Melbourne, Australia, and their caregivers. The present sample comprised of baseline data from children diagnosed with ASD who participated in the evaluation, half of whom were recruited from community football programs, and half who did not participate in organized physical activity but participated in their regular community activities. Participants were recruited through the community football participant database, research registries held by state peak disability bodies, private pediatric clinics, primary schools and special development schools, and social media. To be included in this study, children needed to be aged 5–12 years and have a pre-existing formal diagnosis of ASD. To receive a formal diagnosis of ASD in Victoria, Australia, a child must satisfy diagnostic and statistical manual of mental disorders (DSM) criteria, in which they have undergone assessment by a multidisciplinary panel and have their diagnosis confirmed by a pediatrician or child psychiatrist. Diagnosis was confirmed by caregivers during screening and diagnostic reports were sighted by researchers, where made available by caregivers. Baseline data from the broader study were utilized for the current study.

Measures

Caregivers of participants completed a battery of questionnaires at baseline. Demographic data, including age, gender, and Full-Scale Intelligence Quotient (FSIQ) from age-appropriate Wechsler tests of Intelligence (e.g., Wechsler, 2011, 2012, 2014), were collected. Only the measures that are relevant to this study are reported below.

Participation Environment Measure Children and Youth (PEM-CY; Coster et al., 2011). The PEM-CY community average frequency and average involvement subscales were administered to measure child participation in the community, and the community average perceived environmental barriers and support subscale was administered to measure caregiver perceptions of community-level supports and barriers to their child's participation. The average frequency and involvement subscales consist of 10 items related to activities typically performed in the community, specifically, community events, organized physical activity, unstructured physical activity, classes or lessons outside of school, organizations, clubs, groups or leadership activities, religious activities, "getting together" with other children in the community, and staying overnight (i.e., for a sleepover, holiday or camp). For average frequency, caregivers are asked to indicate how often their child participates on an 8-point scale, with responses including daily, few times a week, once a week, few times a month, once a month, few times in last 4 months, once in last 4 months, or never. For average involvement, caregivers are asked to indicate how involved their child is when participating in these activities on a 5-point scale, with responses ranging from minimally involved to very involved. The community average perceived environmental barriers and support subscale include nine items identifying a number of potential supports and barriers to participation, such as peer relationships, weather conditions and physical layout, and a further seven items identifying community resources. For the first nine items, caregivers were asked to indicate whether the environmental barriers and support made participation easier or harder for their child on a 4-point scale, with possible responses

including "not an issue," "usually helps," "sometimes helps/sometimes makes harder," and "usually makes harder." Three of the resource items were asked to caregivers to indicate whether community resources were available and adequate on a 4-point scale, with possible responses including "not needed," "usually, yes," "sometimes yes/sometimes no," and "usually, no," and the final four items provided a 3-point scale (as per 4-point but with "not needed" removed). Community supportiveness was computed as the average of responses and converted to percentage scores. Items that caregivers indicated a specific barrier or support was not relevant to their child were excluded from the total percentage score. Child participation in community activities was computed as the percentage of activities in which the child participates, with higher scores indicating more activities. Child involvement or engagement in community activities was computed as the average of scores for responses, with higher scores indicating higher levels of engagement. The PEM-CY has demonstrated adequate internal consistency and test–retest reliability (Coster et al., 2011). Due to our small sample size and the volume of "not applicable" responses for this scale, we were unable to accurately determine reliability in our sample.

Vineland Adaptive Behavior Scales, Third Edition (VABS-III; Sparrow et al., 2016). The domain-level parent/caregiver form was administered to measure the adaptive level of functioning in children. The VABS-III domain-level form consists of five domains (communication, daily living skills, socialization, motor skills, and maladaptive behavior), each of which contains 40 items. Raw scores on subdomains of the scale are converted to percentile scores. An overall adaptive behavior composite score is calculated from the communication, daily living, and socialization items (M = 100, SD = 15). Scores of 70 or below reflect a low adaptive level, scores from 71 to 85 reflect moderately low, scores of 86–113 indicate adequate adaptive levels, scores of 115–129 reflect moderately high adaptive levels, and scores of 129 or more indicate a high adaptive level. The VABS-III has demonstrated high test–retest validity and acceptable levels of internal consistency for subdomains (Sparrow et al., 2016). Reliability in our sample was not established.

Parenting Stress Index, Fourth Edition (PSI-IV; Abidin, 2012). The PSI was administered to measure caregiver stress levels in relation to their child and to identify the domains in which these stress may originate from. The PSI asks caregivers to indicate their level of agreement to 101 items using a 4-point Likert scale to measure stressors across three key domains: child factors, caregiver factors, and situational or demographic factors. Scores were summed and percentiles calculated, with higher scores indicating higher stress in that domain. Scores above the 85th percentile on the PSI indicate a clinical level of stress, scores between 81 and 84 are considered high, and scores between 15 and 80 are considered typical levels of stress. The PSI has demonstrated good reliability (Abidin, 2012), with the total PSI score, child domain, and parenting domains demonstrating high reliability (Cronbach's alpha = 0.94, 0.87, and 0.94, respectively), and subscales ranging from 0.63 (acceptability) to 0.89 (spouse/caregiving partner relationship). The life stress subscale did not have adequate reliability (Cronbach's alpha = 0.36) and was not included in analyses.

Social Responsiveness Scale, Second Edition school-aged form (SRS-2; Constantino and Gruber, 2012). The SRS-2 was administered to quantify the severity of ASD symptom in children. This 65-item 4-point Likert scale measures the degree to which caregivers feel each item applied to their child in the preceding 6 months, measuring five areas: social awareness, social cognition, social communication, social motivation, and restricted interests and repetitive behavior. Items were summed, with higher scores indicating more severe deficiencies in symptom severity. Total scores of 76 or more indicate severe deficiencies, scores between 66 and 75 indicate moderate deficiencies, scores between 60 and 65 indicate mild deficiencies, and scores of 59 or less are considered to be within normal limits. The SRS-2 has demonstrated good construct validity and internal consistency with primary-school aged children (Wigham et al., 2012), with the total SRS score and the Social Communication Index demonstrating high reliability (Cronbach's alpha = 0.94 and 0.94, respectively), and subscales ranging from 0.63 (awareness) to 0.88 (Communication) in our sample.

Procedure

Ethical approval was provided by the Deakin Human Research Ethics Committee and the Victorian Department of Education and Training. Those who indicated interest in participating were provided with a plain language statement, and caregivers provided written informed consent while children gave verbal assent. Questionnaires were completed by caregivers while their child participated in testing sessions held at university campuses, football clubs, private clinics and school-based sessions across Victoria as part of the larger longitudinal project, or in some cases questionnaires were completed at home and returned by post.

Statistical Analysis

All statistical analyses were conducted using IBM SPSS statistics version 25. Missing data of less than 5% were treated as per scoring instructions for PSI and, similarly, less than 10% missing data were treated as per scoring instructions for SRS-2. VABS-II does not allow any missing data, and PEM-CY is averaged to account for missing data. For data that exceeded the criterion for missing responses across participants, Little's Missing Completely at Random test indicated that data were missing at random (χ^2 = 307.46, df = 365, p = 0.99) and so list-wise deletion was used. Data were not normally distributed and the sample was small, so two-tailed Kendall's tau correlations were conducted to identify significant relationships between variables. Hierarchical regression was conducted using variables significantly associated with child community involvement, child community attendance, and caregiver stress (isolation). Mahalanobis Distances were calculated and outliers with probability lower than 0.001 were removed. P-P plots were examined to assess whether residuals were normally distributed, and collinearity statistics (VIF and tolerance) and Durbin-Watson statistics were examined. All assumptions were met. Assuming the following parameters – large effect size, α error probability of 0.05, a maximum of six predictors and power of 0.80 – a total sample size of 46 was required.

RESULTS

Participant Characteristics and Correlations

Four female and 44 male children aged 5–12 years (M = 8.41; SD = 2.16) with a full-scale IQ ranging from 41 to 134 (M = 87.8; SD = 20.83) participated. An additional eight participants did not complete the questionnaire items pertaining to the outcome measures, and so were not able to be included in the analyses. Independent t-tests and chi-square analyses did not identify significant differences between those with missing data on outcome measures. Caregivers consisted of 30 mothers and nine fathers, with nine caregivers not reporting their gender. Caregivers' ages ranged from 30 to 52 years (M = 41.40, SD = 5.36). Fourteen caregivers (31%) scored above the 85th percentile on the PSI, indicating a clinical level of stress. All other participants were within the normal range. Means and standard deviations for study variables are found in **Table 1**, as are correlations between variables.

All children with ASD had participated in some kind of neighborhood activity, however, 12% had never participated in a community event, 33% had never participated in organized physical activity, 2% had never participated in unstructured physical activity, 69% had never participated in classes or lessons outside of school, 88% had never participated in organizations, clubs, groups or leadership activities, 63% had never participated in religious activities, 22% had never "gotten together" with other children in the community, and 45% had never stayed overnight (i.e., for a sleepover, holiday, or camp).

Fifty-five percent of caregivers identified features of the environment as not supportive of their child's community participation, and 27% of caregivers felt that information or equipment/supplies at community activities were not adequate for supporting their child's participation. Social demands were identified most frequently as a barrier (35% of caregivers), followed by cognitive demands (33%), sensory demands (22%), physical demands (16%), relationships with peers, attitudes in the community and community safety (8%), and weather conditions (4%).

Child Community Attendance

As shown in **Table 1**, the only significant correlation identified between child characteristics and community participation was child FSIQ (p = 0.004). To explore whether community supportiveness accounts for variations in child community attendance beyond the effects of child characteristics, a stepped multiple linear regression analysis was conducted with FSIQ entered at step 1 and community supportiveness in step 2. As shown in **Table 2**, child FSIQ significantly predicted child community attendance, $F(1,42) = 6.18$, p = 0.02, accounting for 13% of the variance in child community attendance. After controlling for child FSIQ, community supportiveness, entered in step 2, did not significantly predict child community attendance, $F(2,41) = 3.25$, p = 0.05, F-change = 0.40, p = 0.53.

Child Community Involvement

To explore whether community supportiveness accounts for variations in child community involvement beyond the effects of child characteristics, a stepped multiple linear regression analysis was conducted with social awareness, social motivation, restricted interests and repetitive behaviors and adaptive behavior entered at step 1, and community supportiveness in step 2. As shown in **Table 3**, child characteristics revealed a collective effect on child community involvement $F(4,40) = 5.96$, $p = 0.001$, which accounted for 37% of the variation in child community involvement. After controlling for child characteristics, community supportiveness, entered in step 2, further predicted child community involvement, $F(5,39) = 6.21$, $p < 0.001$, F-change = 0.4.90, p = 0.03. The combined predictors accounted for 44% of the variation in child community involvement. The individual predictors were examined further and indicated that adaptive behavior (t = 3.93, $p < 0.001$) and community supportiveness (t = 2.21, p = 0.03) were significant predictors in the model.

Caregiver Stress

Caregiver perceptions of increased overall community support were associated with caregivers perceiving lower levels of isolation (p = 0.001). Correlation analyses, reported in **Table 4**, were run to identify child variables associated with caregiver isolation. Community supportiveness and child characteristics associated with caregiver isolation (child adaptive behavior) were added into a two-step hierarchical regression analysis with caregiver isolation as the criterion variable. The child predictor (adaptive behavior) was added in step 1, and community supportiveness was added in step 2. As shown in **Table 5**, the hierarchical regression model revealed that at step 1 there was a collective significant effect of adaptive behavior $F(1,43) = 4.69$, p = 0.04, with low adaptive behavior explaining 10% of the variance in caregiver isolation. Adding community supportiveness to the model resulted in a significant model $F(2,42) = 8.01$, p = 0.001. Community supportiveness explained 18% of the variance in caregiver isolation, and this change was significant (p = 0.003). The individual predictors were examined further and indicated that adaptive behavior was no longer a significant predictor when community supportiveness was included in the model ($t = -1.45$, p = 0.15), while community supportiveness was a significant predictor in the model ($t = -3.21$, p = 0.003).

DISCUSSION

The current study aimed to address gaps in the literature regarding community inclusion of children with ASD and their families through identifying correlates of child participation and involvement in community activities, caregiver perceptions of community supportiveness, and caregiver stress. Adaptive behavior and ASD symptom severity can impede the ability of children with ASD to meet the various cognitive, social, and communication demands of participation in community activities (Bar-Shalita et al., 2008; Hochhauser and Engel-Yeger, 2010; Obrusnikova and Cavalier, 2011; Poon, 2011; Shattuck et al., 2011; Thompson and Emira, 2011; Krieger et al., 2018; May et al., 2018). Our findings partially

TABLE 1 | Means, standard deviations, and correlations between study variables.

		Community environmental supportiveness (n = 48)	Child community attendance frequency (n = 48)	Child community involvement (n = 48)	Parenting stress isolation (n = 45)
Variable (n)	Mean (SD)	79.08 (11.07)	57.92 (16.24)	3.43 (0.84)	66 (24.93)
Social responsiveness scale					
Social awareness (45)	13.02 (3.31)	−0.21	−0.13	−0.22*	0.08
Social cognition (45)	19.00 (5.90)	−0.19	−0.16	−0.08	0.10
Social communication (45)	33.93 (10.10)	−0.29	−0.12	−0.19	0.21
Social motivation (45)	15.09 (5.53)	−0.15	−0.16	−0.21*	0.05
Restricted interests and repetitive behaviors (45)	20.33 (5.54)	−0.05	−0.14	−0.24*	0.13
Social communication and interaction (45)	81.04 (21.91)	−0.25	−0.11	−0.19	0.17
Total (45)	101.38 (26.26)	−0.22	−0.07	−0.19	0.18
Parenting stress index					
Distractibility (45)	85.44 (15.67)	−0.04	-	-	-
Adaptability (45)	85.93 (14.19)	−0.12	-	-	-
Reinforces caregiver (45)	62.09 (22.75)	−0.13	-	-	-
Demandingness (45)	85.33 (17.65)	−0.17	-	-	-
Mood (45)	79.49 (20.80)	−0.02	-	-	-
Acceptability (45)	82.22 (14.41)	−0.11	-	-	-
Child domain (45)	86.04 (12.45)	−0.15	-	-	-
Competence (45)	61.40 (25.12)	−0.09	-	-	-
Isolation (45)	66.00 (24.93)	−0.36**	-	-	-
Attachment (45)	47.89 (22.14)	0.02	-	-	-
Health (45)	74.29 (21.14)	−0.12	-	-	-
Role restriction (45)	70.89 (25.50)	−0.17	-	-	-
Depression (45)	65.36 (25.96)	−0.10	-	-	-
Spouse/caregiving partner relationship (45)	60.60 (27.50)	−0.10	-	-	-
Caregiving domain (45)	63.78 (22.93)	−0.14	-	-	-
Total (45)	77.29 (14.54)	−0.18	-	-	-
Vinelands adaptive behavior scales					
Adaptive behavior (47)	72.51 (9.60)	0.28	−0.14	0.31**	−0.33**
Communication (47)	75.11 (13.45)	0.18	−0.15	0.25*	−0.25*
Daily living (47)	74.43 (13.14)	0.32*	−0.11	0.31**	−0.25*
Socialization (47)	72.11 (10.11)	0.24	−0.11	0.22*	−0.25*
Motor skills (33)	82.76 (12.79)	0.27	−0.16	0.14	−0.09
Internalizing (46)	19.50 (2.24)	−0.07	−0.07	0.11	−0.01
Externalizing (46)	18.72 (2.65)	−0.33*	−0.03	0.09	0.16
Age of child (48)	8.41 (2.16)	−0.35*	−0.02	−0.10	0.02
FSIQ (44)	87.07 (20.48)	−0.04	−0.30*	0.01	0.10

*Participant numbers varied across measures due to incomplete data. *Significant at 0.05; **Significant at 0.01.*

TABLE 2 | Predicting child community participation from child IQ and community supportiveness.

Variable	*B*	*β*	*p*	sr^2	*R*	R^2	*F*	*Sig.*
Step One					0.36	0.13	6.18	0.02
(constant)	5.11	-	<0.00	-				
FSIQ	−0.01	−0.36	0.02	−0.36				
Step Two					0.37	0.14	3.25	0.05
(constant)	4.53	-	<0.00	-				
FSIQ	−0.01	−0.35	0.02	−0.35				
Community supportiveness	0.01	0.09	0.53	0.10				

supported this former research by showing that community attendance and involvement rates of children with ASD were low and significantly associated with FSIQ (attendance) and adaptive behaviors and ASD symptom severity, such as social awareness, social motivation, and restricted interests and repetitive behaviors (involvement).

The relationships between child characteristics and child involvement were stronger than those with child attendance, indicating that child adaptive behaviors and ASD symptom severity may be more important for child engagement and involvement in community programs than for the attendance of children with ASD in community programs. Specifically, greater ASD symptom severity and fewer adaptive behaviors are linked to lower quality of community engagement but not lower quantity. The findings that FSIQ predicted child community attendance aligns with previous research finding the cognitive demands of community activities act as a barrier for children with ASD (Egilson et al., 2017); however, the lack of other significant predictors for child community attendance was unexpected.

TABLE 3 | Predicting child community involvement from child social responsiveness, adaptive behavior and restricted interests and repetitive behaviors.

Variable	*B*	*β*	*p*	sr^2	*R*	R^2	*F*	*Sig.*
Step One					0.61	0.37	5.96	0.001
(constant)	0.14	-	0.94	-				
Social awareness	0.02	0.24	0.17	0.22				
Social motivation	−0.01	−0.13	0.44	−0.12				
Restricted interests and repetitive behaviors	−0.02	−0.18	0.29	−0.17				
Adaptive behavior	0.05	0.56	<0.001	0.53				
Step Two					0.67	0.44	6.21	<0.001
(constant)	−1.21	-	0.51	-				
Social awareness	0.02	0.28	0.10	0.26				
Social motivation	−0.01	−0.11	0.50	−0.11				
Restricted interests and repetitive behaviors	−0.02	−0.22	0.19	−0.21				
Adaptive behavior	0.05	0.50	0.001	0.50				
Community supportiveness	0.02	0.28	0.03	0.33				

TABLE 4 | Correlations between caregiver isolation and child variables.

Variable (*n*)	Parenting stress isolation (*n* = 45)
Social responsiveness scale	
Social awareness (45)	0.08
Social cognition (45)	0.10
Social communication (45)	0.21
Social motivation (45)	0.05
Restricted interests and repetitive behaviors (45)	0.13
Social communication and interaction (45)	0.17
Total (45)	0.18
Vinelands adaptive behavior scales	
Adaptive behavior (47)	−0.33**
Communication (47)	−0.25*
Daily living (47)	−0.25*
Socialization (47)	−0.25*
Motor skills (33)	−0.09
Internalizing (46)	−0.01
Externalizing (46)	0.16
Age of child (48)	0.02
FSIQ (44)	0.10

*Significant at 0.05; **Significant at 0.01.

Previous research has shown a range of links for child attendance with child characteristics and community supportiveness (Egilson et al., 2017; Krieger et al., 2018), and the current findings conflict with this former literature.

In line with the social model of disability, our findings highlighted the role community supportiveness plays in child involvement in community activities. As expected, a high number of caregivers indicated community environments did not support their child's participation. Regardless of a child's symptom severity, community supportiveness significantly predicted child involvement, supporting the premise that children with ASD can experience successful involvement in their community with the right supports in place, or conversely, experience poor involvement when their community is not supportive. These findings fit with previous research indicating community factors, such as poor predictability, social and cognitive demands and low availability, and suitability of community services and staff (Egilson et al., 2017; Krieger et al., 2018), can disrupt child participation, and extend these findings through demonstrating that community supportiveness may be more important for involvement than the child characteristics themselves. These findings provide promising initial support that adapting community programs and activities for children with ASD may lead to increased involvement of these children. Of note, however, reduced child adaptive behavior significantly predicted child involvement irrespective of community supportiveness. This may indicate an area in which intensive supports and intervention may be needed to increase the ability of children with ASD to be fully involved in their community.

Caregiver Stress

Caregiver stress in our sample was high, with one in three caregivers scoring above the clinical cut-off on the Parenting Stress Index. Previous research found caregivers who perceive social support received by themselves or their child as inadequate are more likely to experience high levels of stress (Siklos and Kerns, 2006; Siman-Tov and Kaniel, 2011), and our results suggest that caregivers of children with ASD who perceive their community as being unsupportive of their child's participation in the community due to the presence of many barriers, low levels of helpfulness, and few resources available may experience higher levels of isolation. Similar to research of Hall (2012), in which child ASD symptom severity and community supportiveness accounted for 16% of the variance in family coping scores, community supportiveness explained 18% of the variance in caregiver isolation in the current study. Community supportiveness was not linked to other forms of stress in our sample. This may indicate that where other studies found other forms of low social supports (i.e., friends and family) were linked to high caregiver stress (Hall, 2012), community supportiveness may be less important for other forms of stress beyond feelings of isolation.

Of particular note is the finding that when variations in caregiver perceptions of community supportiveness were accounted for, child adaptive behaviors no longer significantly predict caregiver isolation. These findings suggest that low levels of community supports for child participation contribute more strongly to caregiver isolation than child functioning does, indicating that increasing community supportiveness of children with ASD may also decrease caregiver isolation. This fits with previous qualitative research in which caregivers indicated low levels of community support resulted in caregivers taking on an advocacy role for their child, which left them feeling labeled and isolated (Resch et al., 2010).

Limitations

The research findings need to be interpreted in the context of a number of limitations. First, given the current study was limited to cross-sectional data, directionality cannot be confirmed. In particular, it is unclear at this stage whether caregivers who feel isolated are more likely to perceive increased barriers to their child's participation, or whether barriers to

TABLE 5 | Predicting parenting isolation from child adaptive behavior and community supportiveness.

Variable	*B*	*β*	*p*	sr^2	*R*	R^2	*F*	*Sig.*
Step One					0.31	0.10	4.69	0.04
(constant)	128.67	-	0.00	-				
Adaptive behavior	−0.86	−0.31	0.04	−0.31				
Step Two					0.53	0.28	8.01	0.001
(constant)	184.57	-	0.00	-				
Adaptive behavior	−0.55	−0.20	0.15	−0.22				
Community supportiveness	−0.98	−0.44	0.003	−0.44				

child participation lead to feelings of isolation in caregivers. It is also possible that this relationship is reciprocal, with increased barriers leading to feelings of isolation, and increased feelings of isolation leading to an increasingly negative view of community supportiveness for child inclusion; however, further research is needed to establish directionality. Second, Wechsler tests of Intelligence can underestimate cognitive functioning. This study's aim was to control variations in FSIQ and, therefore, this underestimation is unlikely to impact results; however, future research could consider examining other non-verbal measures of cognitive functioning in relation to community participation. Finally, the sample size for this study was small and, therefore, only powered to detect large effects, consisted of caregivers of predominantly male children, and half of the sample consisted of children with autism recruited from a community football program. Further, reliability of PEM-CY and Vinelands was not established for our sample, and variations in reliability have been noted for the PEM-CY previously (see Coster et al., 2011; Simpson et al., 2019). Replication in a larger and more diverse population would address these limitations.

CONCLUSION

In summary, these findings suggest that lower perceived levels of community supportiveness may reduce the involvement of children with ASD in community activities and increase feelings of isolation in their caregivers. Specifically, children with ASD may experience increased inclusion in programs that cater for varying communication, cognitive and social abilities, and in addressing key barriers to participation of children, caregivers may experience reduced feelings of isolation. Disruptions to adaptive behaviors in children with ASD may pose particular challenges for children with ASD, and further research exploring intensive intervention and supports is warranted.

The findings that child characteristics and community supportiveness may have more impact on child involvement or engagement in community activities than on attendance raise questions as to whether reduced quality of engagement in community activities in children with ASD disrupts the benefits of regular participation. Future research delineating participation and involvement could explore if low engagement or involvement in community activities results in lower levels of beneficial outcomes in children with ASD despite regular participation, and test whether specific program attributes related to accessibility and inclusivity impact child involvement. Further research is also needed to identify and evaluate the effective modifications to community programs in these key areas and to measure their collective impact on caregiver perceptions of community supportiveness and caregiver isolation. Community sports that are tailored to facilitate the inclusion of children with ASD have been increasingly recognized over the past decade as a promising intervention medium (Rinehart et al., 2018; Howells et al., 2019). The findings that adaptive behavior and community supportiveness support child involvement in community settings play an important role in providing an evidence-based approach for inclusion in community sports settings, while seeking to ensure that the benefits of these approaches extend beyond merely boosting attendance of a community program, and instead reflect a deeper engagement and connection within these settings. This study provides clear insights into the potential for inclusive and adapted community programs to facilitate active engagement and participation in children with ASD, while reducing isolation stress in caregivers.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Deakin University Human Research Ethics Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

All authors were involved in data interpretation and manuscript drafting. All authors read and approved the final manuscript.

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Social Cognition, Social Skill, and Social Motivation Minimally Predict Social Interaction Outcomes for Autistic and Non-Autistic Adults

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Social cognition, social skill, and social motivation have been extensively researched and characterized as atypical in autistic people, with the assumption that each mechanistically contributes to the broader social interaction difficulties that diagnostically define the condition. Despite this assumption, research has not directly assessed whether or how these three social domains contribute to actual real-world social interaction outcomes for autistic people. The current study administered standardized measures of social cognition, social skill, and social motivation to 67 autistic and 58 non-autistic (NA) adults and assessed whether performance on these measures, both individually and relationally between dyadic partners, predicted outcomes for autistic and NA adults interacting with unfamiliar autistic and NA partners in a 5 minute unstructured "get to know you" conversation. Consistent with previous research, autistic adults scored lower than NA adults on the three social domains and were evaluated less favorably by their conversation partners. However, links between autistic adults' performance on the three social domains and their social interaction outcomes were minimal and, contrary to prediction, only the social abilities of NA adults predicted some interaction outcomes within mixed diagnostic dyads. Collectively, results suggest that reduced performance by autistic adults on standardized measures of social cognition, social skill, and social motivation do not correspond in clear and predictable ways with their real-world social interaction outcomes. They also highlight the need for the development and validation of more ecological assessments of autistic social abilities and the consideration of relational dynamics, not just individual characteristics, when assessing social disability in autism.

Keywords: social interaction, social cognition, social skills, first impressions, double empathy

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INTRODUCTION

Autism Spectrum Disorder (ASD) is clinically defined in part by "persistent deficits in social communication and social interaction" (APA, 2013). Although the focus on communication and interaction necessitates consideration of interpersonal and relational dynamics, the focus on deficits—or reductions in normative characteristics presumed to underlie autistic disability—has historically centered research and treatment at the level of the individual. Indeed, a deep literature has accumulated cataloging how autistic people differ from non-autistic (NA) people in

their neurology, cognition, and behavior (Pelphrey et al., 2004; Chevallier et al., 2012a; Morrison et al., 2017, 2019b), and a wide variety of intervention programs have been developed using this evidence base to try to normalize individual characteristics with the presumption that doing so may reduce or mitigate autistic disability (for a review, see Pallathra et al., 2019). For autistic adults without intellectual disability, most of these programs are psychosocial in nature and target three primary domains of social ability: social cognition, social skill, and social motivation. Each has been characterized as a core autistic deficit and are assumed to underlie the occupational challenges (Taylor et al., 2015), social isolation (Mazurek, 2014), and reduced quality of life (Billstedt et al., 2005) often experienced by autistic adults.

Social cognition refers to the perception and interpretation of social information (Brothers, 1990) and is often conceptualized as encompassing social perception (i.e., the prioritization and detection of social information), emotion recognition (i.e., accurately identifying the emotional state of others), and theory of mind (i.e., inferring the thoughts and intentions of other people; Baron-Cohen, 1991; Happé, 1994; Mathersul et al., 2013). On average, autistic adults score lower than NA controls on standalone assessments on each subdomain (Morrison et al., 2019b). They score lower on tasks assessing face recognition (e.g., Klin et al., 1999; Joseph and Tanaka, 2003), the identification of emotion from facial expressions, voices, and social scenes (e.g., Golan et al., 2007; Kennedy and Adolphs, 2012; Uljarevic and Hamilton, 2013; Sasson et al., 2016), and the inference of other peoples' intentions and mental states (e.g., Spek et al., 2010; Mathersul et al., 2013). Although these difficulties are presumed to mechanistically relate to the poor social and functional outcomes autistic adults often experience (Sasson et al., 2011), the surprisingly small number of studies that have empirically tested this assumption tend to find only modest relationships (Klin et al., 2002; Lerner and Mikami, 2012; Bishop-Fitzpatrick et al., 2014; Hanley et al., 2014; Deschrijver et al., 2016; Sasson et al., 2020), and no studies to our knowledge have tested whether individual social cognitive performance demonstrates meaningful associations to real-world social interaction for autistic adults. Given that social cognition is often targeted for improvement in psychosocial interventions as a means for enhancing social interaction, the lack of evidence in this regard reflects a significant oversight.

Social skill, meanwhile, is a broad umbrella term referring to the repertoire of behaviors used to navigate social demands and achieve social goals across varying contexts (Mueser and Bellack, 1998). A diverse set of skills have been conceptualized to comprise social skill, ranging from the use of interpersonal eye gaze to more complex competencies like negotiation ability (Mueser and Bellack, 1998; Nangle et al., 2010). Social skills reliably differ in autism (Constantino et al., 2000; Hus and Lord, 2014), with autistic adults often exhibiting non-normative social behaviors within social interactions relative to NA controls (Bishop, 1998; Patterson et al., 2001; Verhoeven et al., 2013). These differences can include atypical use of gaze, less observable conversational involvement, reduced verbal fluency, atypical affect, and asking fewer questions of their interaction partner (Ratto et al., 2011; Morrison et al., 2017). Training programs targeting social skills to improve social functioning among autistic adolescents and adults have yielded some limited benefits (Wykes et al., 2008; Bishop-Fitzpatrick et al., 2014), but they tend to lack generalizability to real-world outcomes (Palmen et al., 2010; Gates et al., 2017; Bottema-Beutel et al., 2018). This may stem in part from an overreliance on examining autistic social skill as an isolated, individual characteristic rather than assessing how it manifests within the context of actual interaction in which relational dynamics and not just individual behavior dictate outcomes (Milton, 2012; Bolis et al., 2018). It also suggests that a single, normative standard for social skill may not conform equally to the communication preferences and expectations of all groups or individuals. Indeed, this criticism is central to the Double Empathy theory of autism (Milton, 2012), which argues that social barriers between autistic and NA people are not solely driven by autistic misunderstanding of NA communication and behavior (as commonly described within autism research) but also the reverse: NA people often exhibit difficulty inferring the mental states and interpreting the social cues of autistic people (Edey et al., 2016; Alkhaldi et al., 2019). From this perspective, social skill is relative, contextual, and necessitates a focus on relational dynamics rather than individual ability.

Finally, social motivation refers to the seeking and liking of social information and relationships (Berridge et al., 2009; Chevallier et al., 2012a). Young children on the autism spectrum often demonstrate reduced attention and divergent reward responses for social information from early in life (Baranek, 1999; Pierce et al., 2011; Chevallier et al., 2012a; Moriuchi et al., 2017), which is theorized to produce cascading effects on developing social neural networks that manifest over time in divergent social behaviors and social cognitive abilities relative to same age peers (Dawson et al., 2005; Chevallier et al., 2012a). In older autistic children and adolescents, some work suggests that diminished social motivation may result in fewer social exchanges and less effort toward maintaining relationships (e.g., Chevallier et al., 2012b). However, many other studies have found that social motivation is highly variable among autistic adolescents and adults (Garman et al., 2016), most of whom express similar desires for friendships and relationships as their NA peers (Bauminger and Kasari, 2000; Whitehouse et al., 2009; Lasgaard et al., 2010; Mazurek, 2014). Higher social motivation among autistic individuals may relate to having better quality friendships, engaging in more social interactions, and displaying higher rates of prosocial behavior in interactions with others (Chevallier et al., 2012b; Dean et al., 2014; Sedgewick et al., 2016). At the same time, lower or different social interest is not inherently negative (Dawson and Cowen, 2019; Fletcher-Watson and Crompton, 2019), and pressure to conform to normative expectations can be detrimental to autistic well-being (Cage and Troxell-Whitman, 2019). For instance, many autistic adolescents and adults without intellectual disability report engaging in effortful and often exhausting "camouflaging" behaviors in order to appear more typical within social interactions (Hull et al., 2017). These deliberate masking behaviors suggest that—rather than lacking motivation for relationships—autistic individuals may instead differ in their social skill and communication styles, struggle to have their social needs met, and expend tremendous

effort trying to fit in (Hintzen et al., 2010; Chevallier et al., 2012b; Garman et al., 2016).

Collectively, this broad body of research has delineated reliable group-level differences between autistic and NA people on a range of social cognitive, social skill, and social motivation measures, with findings more variable and idiosyncratic concerning their association with social and functional outcomes. One potential reason for the lack of more established links with broader outcomes is that no studies have assessed whether and how these three social abilities relate to social interaction success or difficulties for autistic adults. Social interaction serves as the interface between the individual abilities and social outcomes, and despite social interaction difficulties constituting a core diagnostic component of autism, no research to our knowledge has systematically examined whether social cognition, social skill, and social motivation correspond to real-world social interaction outcomes for autistic adults. Critically, social interaction involves more than one person and necessitates consideration of not just an individual's social abilities, but also those of the interaction partner—and the relational combination between them—in order to understand how each partner influences the other (De Jaegher, 2013; Hehman et al., 2017). Research in autism, however, has focused overwhelmingly at the level of the individual, with studies of social interaction even being called a "blind spot" (De Jaegher, 2013) because so few studies have assessed dynamic interaction amongst and between autistic people.

This has started to change in recent years. Recent empirical work with autistic adults has shown that social interaction quality and positive perceptions are driven by relational factors to a greater degree than individual ones (Crompton et al., 2020; Morrison et al., 2020). For instance, autistic adults disclose more about themselves (Morrison et al., 2020), communicate more effectively, and establish better rapport (Crompton et al., 2020) when interacting with other autistic adults relative to NA adults. This suggests that relational compatibility, and not just individual characteristics, contribute to social interaction outcomes for autistic adults, but it remains unclear whether specific social abilities either individually or dyadically underlie this effect.

A previous study (Morrison et al., 2020) reported that autistic adults were evaluated less favorably by both autistic and NA partners after engaging in a real-world "get to know you" conversation, and NA adults expressed a preference for future social interaction with NA relative to autistic adults. In contrast, autistic adults trended toward preferring interaction with autistic relative to NA adults. The current study analyzes additional data from this sample to assess whether and how three aspects of social ability (i.e., social cognition, social skills, and social motivation) relate to social interaction outcomes for autistic and non-autistic adults across three dyadic combinations of diagnostic status (i.e., A-A, NA-NA, A-NA). The Actor-Partner Interdependence Model (APIM; Kenny et al., 2006) was used to assess the effect of individuals' social abilities on their own evaluations of their partner and the interaction (actor effects), the effect of the partners' social abilities on how individuals evaluate that partner and the interaction (partner effects), and the interaction between the two (actor-partner interactions). We predicted that (1) regardless of diagnosis, individuals with lower social cognitive performance, social motivation, and observed social skill will evaluate their partner less favorably and rate their own experience of the interactions lower in quality and closeness (i.e., actor effects); (2) regardless of diagnosis, individuals with lower social cognitive performance, social motivation, and observed social skill will be evaluated less favorably by their partners and their partners would rate their experience of the interactions lower in quality and closeness (i.e., partner effects); (3) actor, partner, and actor-partner interaction effects involving social variables will be moderated by diagnosis, such that effects of social abilities on outcomes will be stronger for autistic compared to NA individuals; and (4) actor, partner, and actor-partner interaction effects involving social variables will be moderated by dyad type, such that the effect of social abilities on outcomes will differ depending on whether dyads share or differ in their diagnostic status. Together, these hypotheses assess which factors predict person and interaction evaluations, and what combinations of partners and/or traits lead to poor or favorable interactions.

METHODS

Participants

One hundred and twenty-five adults (67 A, 58 NA) participated in one of three types of conversation dyads: A-A ($n = 22$), A-NA ($n = 25$), and NA-NA ($n = 23$). Autistic participants were recruited from the UT Dallas Autism Research Collaborative, a research registry of clinically assessed autistic adults who have expressed interest in participating in university research studies. Inclusion in the registry requires confirmed diagnoses using the ADOS-II (Lord et al., 2000) and full-scale intelligent quotients (IQ) over 70 on the WASI-II (Wechsler, 2011), both of which occurred during a previous clinical intake session. Full-scale IQs over 90 were required for this study in order to be intellectually comparable to the NA participants. All included NA participants were university undergraduates, and only those who reported no history of psychiatric illness (8 excluded), no developmental disability (including autism; 1 excluded), and no autistic first-degree relatives (4 excluded) were retained in the study. Those with autistic first-degree relatives were excluded to minimize inclusion of NA adults with high familiarity with autism and/or autistic traits. Additionally, two autistic adults were excluded for having an IQ lower than 70. The protocol for the study was approved by the University Institutional Review Board, and all participants provided informed consent before the study began.

Autistic and NA participants were recruited with the intent of matching the diagnostic groups and the three dyad types demographically. All participants were male to avoid the complicating dynamics of inter-sex dyads and because the higher male ratio in autism (Fombonne, 2009) and in our recruitment sources precluded a well-powered examination of gender effects. Autistic and NA participants differed in age (A mean = 23.51, SD = 4.07; NA mean = 20.84, SD = 3.17; $p < 0.01$) but did not differ on race (A = 84% White; NA = 81% White; $p = 0.83$) and estimated IQ on the Wide Range Achievement Test (WRAT-3; Wilkinson, 1993; A mean = 110.77, SD = 8.58; NA mean = 109.91, SD = 8.39; $p = 0.58$), a brief assessment that correlates highly with full scale IQ (Powell et al., 2002). The three dyad

types did not differ on race ($p = 0.97$) or estimated IQ ($p = 0.17$), but did on age ($p < 0.01$), with the NA-NA group consisting of younger participants than the other two dyad types. To ensure that any findings between diagnostic groups and dyad types were not influenced by demographic characteristics, age, race, and IQ were covaried in all analyses. Demographic characteristics for the diagnostic and dyad groups can be viewed in **Table 1**. For more details about the sample, including information about the descriptive and psychometric properties of all included measures, see Morrison et al. (2020).

Procedure

Potential participants were initially screened for inclusion criteria, scheduling availability, and demographic characteristics. This information was then used to recruit unfamiliar dyadic partnerships of participants similar on age and race. Efforts to recruit unfamiliar conversation partners were largely successful: only one dyad consisted of partners who mutually acknowledged seeing their conversation partner previously, but both said that they had never spoken.

After providing informed consent, participants sat in chairs facing each other and were videotaped while completing an unstructured conversation developed to evaluate dyadic interaction (Berry and Hansen, 1996). This measure originally was created to measure interactions between two NA participants but recently similar paradigms have also been used with autistic adults (Usher et al., 2018; Morrison et al., 2020). Participants are told that they will be talking to their partner for 5 min. No specific instructions are given other than telling them that their goal is to get to know the other person. After instructions were given, the experimenter moved behind a partition to avoid influencing the interaction. Participants were not explicitly made aware of the diagnostic status of their partners, but disclosure occurred organically during interactions within three A-A dyads and three A-NA dyads. Following the conversation, participants completed measures on separate computer stations that (1) recorded their impressions of the interaction and their conversation partner, (2) assessed their social cognitive performance, and (3) measured their social motivation. To ensure order effects did not influence results, these groups of measures were counterbalanced for each participant, and the order of the measures within each group of measures was randomized.

Measures

Evaluation of the Partner and the Interaction

Participants evaluated their conversation partner using the First *Impression Scale for Autism* (FIS; Sasson et al., 2017) and *the International Personality Item Pool—Interpersonal Circumplex (IPIP-IPC;* Markey and Markey, 2007) The FIS includes 10 items using a four-point scale. Participants rated their partner on six traits (awkwardness, attractiveness, trustworthiness, likability, dominance, and intelligence), and completed four items concerning their interest in socializing with their partner in the future (e.g., "I would hang out with this person in my free time"). Because the social interest items, but not the trait items, showed relatively high internal consistency (see Morrison et al., 2020 for details), a composite score averaging the four social interest items was used in analyses, whereas the six trait items were individually included.

The IPIP-IPC consists of 32 items assessing social behavioral characteristics unassessed by the FIS. Specifically, the IPIP-IPC measures interpersonal warmth and dominance, two key predictors of dyadic behavior in social interactions and a variety of interaction outcomes [e.g., relationship satisfaction, task productivity, and liking (Markey and Markey, 2007; Markey et al., 2010)]. Items are aggregated to calculate separate warmth and dominance scores that are then used in analyses.

Participants evaluated qualities of the interaction using *the Social Interaction Evaluation Measure* (SIEM: Berry and Hansen, 1996), the *Inclusion of Other in the Self* (IOS Scale; Aron et al., 1997), and the *Subjective Closeness Index* (SCI; Berscheid et al., 1989). The SIEM is a self-report measure consisting of 11 questions rated on an eight-point scale concerning the participant's perceptions of the interaction quality, the intimacy of the interaction, the partner's level of disclosure, and the partner's level of engagement in the conversation. The items are averaged to create a composite score indicating overall interaction quality (Heerey and Kring, 2007).

The IOS and SCI are measures of interpersonal closeness that are averaged together to create an overall closeness composite score (Aron et al., 1997). The IOS requires the participant to select one of seven overlapping circles that best represents how close they feel to their conversation partner. The SCI uses a seven-point scale to ask the participant to rate their level of closeness to their partner relative to their other relationships and their perception of closeness in the relationships of other people.

Evaluation of Social Abilities: Social Cognition, Social Motivation, and Social Skills

Social cognition

Participants completed three measures spanning the separate domains of social cognition: face perception (Benton Facial Recognition Task; Benton et al., 1983), emotion recognition (Penn Emotion Recognition Task, ER-40; Kohler et al., 2000), and theory of mind (The Awareness of Social Inference Task. TASIT; McDonald et al., 2003). In the Benton, participants view 54 faces and select the matching face from an array of six faces. In the ER-40, participants select one of five emotion choices corresponding to the emotion expressed in 40 face photos. In the TASIT, participants watch 16 short videos depicting characters lying or being sarcastic within social interactions and answer four questions after each video regarding what the characters' intentions, thoughts, and beliefs were about the other people or the scenario. All three tasks have been used in previous studies of autism (Philip et al., 2010; Neves et al., 2011; Ratto et al., 2011) and have been psychometrically validated for inclusion in autism research (Morrison et al., 2019b). As has been done previously (Sasson et al., 2013), social cognitive scores from these three domains were standardized and averaged together to yield a total social cognition composite score used in primary analyses. The independent impact of each social cognition domain on social interaction outcomes was also pursued in exploratory analyses.

TABLE 1 | Demographic characteristics for diagnostic and dyad groups.

	Dyad groups								Overall			
	A-A		NA-NA		A-NA				A		NA	
	(*n* = 42)		(*n* = 40)		(*n* = 42)				(*n* = 66)		(*n* = 58)	
Race												
White	36		33		34				56		47	
Black	2		2		2				3		3	
Asian	2		1		2				3		2	
Other	2		4		4				4		6	
					A		NA					
	M	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
Age	22.67	3.62	20.62	3.43	25.10	4.47	21.33	2.50	23.51	4.07	20.84	3.17
WRAT-3 IQ	111.88	7.12	110.78	7.91	108.67	10.72	108.00	9.34	110.77	8.58	109.91	8.39

WRAT-3, Wide Range Achievement Test 3rd Edition.

Social motivation

Participants completed the Friendship Motivation Scale (Richard and Schneider, 2005) to assess their interest in forming social relationships. Participants answer 12 questions on a 4-point scale across four subscales: intrinsic motivation, identified regulation, external regulation, and amotivation. Intrinsic motivation refers to self-determination for seeking friendships, in which social relationships are satisfying for internal reasons (e.g., for the pleasure I get by talking with friends). The other three types of motivation are extrinsic in nature. Identified regulation refers to seeking relationships for their own sake (e.g., because I think having friends is good for me). External regulation refers to seeking friendships for environmental reasons or rewards (e.g., to be invited to parties). Lastly, amotivation refers to a lack of motivation to seek friendships because the individual does not perceive benefits from friendships (e.g., I don't see why I would want to have friends). The total social motivation score was computed by summing weighted subscale scores, with higher scores signifying higher social motivation (see Richard and Schneider, 2005 for formula).

Social skills

To obtain a measure of both partners' social skills, three independent raters (one autistic) were trained on the Conversation Probe (CP) social behavior coding manual (Pinkham and Penn, 2006). Prior to coding, raters attended training sessions and coded videos until consensus in ratings was achieved on 20% of the videos. All raters were blind to participant diagnoses. The CP captures both discrete social skill ratings and a holistic rating of the participant's overall social skill. Coders first coded nine discrete behaviors categorized into four composite skill groups: appropriate content, paralinguistic behaviors, interactive behaviors, and non-verbal behaviors (Morrison et al., 2017). Conversational content refers to the participant's ability to discuss topics appropriate to meeting someone for the first time. Paralinguistic behaviors quantify the quality of participants' speech other than semantic content (e.g., speaking with clarity, enunciating clearly and fluently, and successfully switching turns with their partner). Interactive behaviors measure the degree to which participants are interested in getting to know their partners and carry on the interaction. This subscale was comprised of involvement, or the degree to which the participants appear engaged in the conversation, and the number of questions the participants asked of their partner. Lastly, non-verbal behaviors consisted of the degree of appropriate eye-contact and affective behaviors displayed by the participants. These social behaviors were originally derived based upon non-autistic norms, and thus the CP should be understood as measuring social skills considered normative and valued by non-autistic society.

Each social skill rating was made on a nine-point Likert scale, where higher scores indicated better social skills ability. Coders also make a holistic judgement of the participant's overall skill ability, rating how successful the participant was at interacting with his partner. Intraclass correlation coefficients (ICCs) were computed to assess reliability on the videos. The three coders' consistency ranged from 0.57 to 0.95 on the behaviors across the full sample of videos and they were strongly consistent for overall social skills (ICC = 0.732). Reliability is displayed in **Table 2**.

Analysis Plan

Before proceeding to our primary analyses, we inspected the descriptive statistics for the study variables and tested whether autistic and NA adults significantly differed in their respective group means. We then investigated the pattern of zero-order correlations between the study variables for autistic and NA adults separately to gain some preliminary insights into possible group differences in the predictor-predictor and predictor-outcome associations.

Because outcomes for partners were interrelated and thus non-independent, traditional analytic techniques (e.g., general linear model) could not be used for primary analyses.

TABLE 2 | Means and group comparison of social skills.

	ICC	A		NA		$F_{(1, 123)}$	p
		M	*SD*	*M*	*SD*		
Content	0.731	6.72	0.81	7.09	0.60	8.177	0.005
Clarity	0.588	6.08	0.96	6.42	0.72	4.934	0.028
Fluency	0.765	6.01	1.08	6.60	0.59	13.817	<0.001
Meshing	0.713	6.02	1.16	6.59	0.68	10.841	0.001
Gaze	0.660	6.67	1.12	7.55	0.57	29.398	<0.001
Involvement	0.793	6.59	1.09	7.21	0.55	15.149	<0.001
Asks questions	0.951	3.76	2.66	5.61	2.42	16.297	<0.001
Appropriate affect	0.655	6.80	0.54	7.12	0.46	12.361	0.001
Flat affect	0.712	5.85	1.00	6.27	0.82	6.548	0.012
Social anxiety	0.725	6.01	0.97	6.78	0.64	27.012	<0.001
Overall skill	0.732	5.57	1.04	6.44	0.62	31.494	<0.001
Repetitive verbal content	0.566	6.79	0.77	7.18	0.42	11.688	0.001
Repetitive movement	0.743	6.56	0.97	7.17	0.55	17.813	<0.001
Verbosity	0.905	6.36	1.85	6.40	1.13	0.022	0.882
Paralinguistic	–	6.04	0.86	6.54	0.47	15.675	<0.001
Non-verbal	–	6.44	0.69	6.98	0.47	25.459	<0.001
Interactive	–	5.18	1.60	6.41	1.34	21.373	<0.001

ICC refers to Intraclass correlation coefficient for coders' reliability. Note the paralinguistic, non-verbal, and interactive behaviors are composite scores rather than coded items, and thus do not have ICCs.

Instead, the Actor-Partner Interdependence Model (APIM) for indistinguishable dyads was used (Kenny et al., 2006). The APIM provides estimates of actor effects (e.g., the effect of individuals' social abilities on their own interaction outcomes), partner effects (e.g., the effect of individuals' partners' social abilities on individuals' interaction outcomes), and (if researchers are interested) actor-partner interactions (e.g., how the effect of individuals' social abilities on their own interaction outcomes depends upon their partners' social abilities). Additionally, by collecting dyads that differed in their diagnostic composition, we could investigate whether effects differed for autistic adults compared to NA adults, as well as whether the particular combination of dyad members (i.e., A-A, A-NA, NA-NA) moderated any effects (Kraemer and Jacklin, 1979; Kenny et al., 1988). **Figure 1** visually displays the model used for analysis.

APIMs were specified using multilevel modeling with Restricted Maximum Likelihood estimation in SPSS Version 25. Multilevel modeling is appropriate because participants and their interaction partners are nested within dyads. It also helps to account for missing data in the outcomes, which were minimal in this study. Actor IQ, race, and age were entered as co-variates in all analyses. To facilitate the interpretation of the coefficients in each of the APIM analyses, continuous predictors were grand-mean centered and categorical predictors were effects coded (moreover, interaction terms assessing moderation were specified using these centered and effect-coded variables). An adjusted alpha of 0.01 was used as the threshold for statistical significance given the large number of tests; however, a more lenient alpha of 0.05 was used when significant interaction terms were followed up to increase our power to detect simple slopes once our more conservative threshold for detecting an interaction was reached.

RESULTS

Descriptive Statistics

Normality, skew, and kurtosis were within acceptable ranges for analyses. Skew and kurtosis values were below the absolute value of 2 for all measures, with two item-level exceptions: aggression/dominance and the behavioral intent composite from the first impression scale exceeded the kurtosis threshold but were still relatively normal in their distributions. Means and standard deviations for social cognitive tasks, social motivation, and social skills can be viewed in **Tables 2**, **3** [those for outcome measures (i.e., first impression scale, IPC warmth, IPC dominance, interaction quality, and closeness) appear in Morrison et al., 2020]. NA adults scored higher than autistic adults on the Benton [$F_{(1, 177)} = 26.37$, $p < 0.001$], TASIT [$F_{(1, 117)} = 14.98$, $p < 0.001$], FMS [$F_{(1, 117)} = 12.46$, $p = 0.001$], social cognition composite score [$F_{(1, 117)} = 26.02$, $p < 0.001$], overall social skills ratings [$F_{(1, 123)} = 31.49$, $p < 0.001$], but diagnostic groups did not differ on the ER-40 [$F_{(1, 117)} = 2.79$, $p = 0.10$].

Social ability predictors were weakly to moderately correlated with one another (**Table 4**). Correlations between predictors and actor and partner outcomes showed that for autistic adults, several social abilities moderately predicted partner evaluations (**Tables 5**, **6**). As can be seen in **Table 5**, for autistic adults, higher social motivation on the FMS was related to perceiving the partner as warmer, less aggressive/dominant, smarter, and having a stronger desire to have a conversation with their partner. ER-40 scores predicted stronger acceptance of living near the partner, but higher theory of mind performance on the TASIT was related to feeling less close to partners, perceiving the partner as less dominant, and having less desire hang out with their partners later. For NA adults, higher ER-40 scores predicted rating partners lower in warmth, and higher TASIT scores were related to perceiving the partner as less dominant and more trustworthy. Higher social motivation on the FMS was related to seeing the partner as more attractive. As can be seen in **Table 6**, autistic adults with better observed social skills were rated as less awkward, smarter, and having higher quality interactions, those with better theory of mind performance on the TASIT were rated as smarter, and those with higher emotion recognition scores on the ER-40 were rated less dominant and more awkward. For NA adults, those with higher Benton facial recognition scores were rated as warmer.

Actor-Partner Interdependence Model (APIM) Analyses

An initial model, detailed in Morrison et al. (2020), was run to assess the effect of the diagnostic status (A or NA) of the actor, partner, and the interaction between them on reports of social interaction quality, closeness, and first impressions of various traits. For the current study, this model was run with additional parameters to examine (a) if social abilities (i.e., social cognition, social motivation, and social skill) predicted social interaction

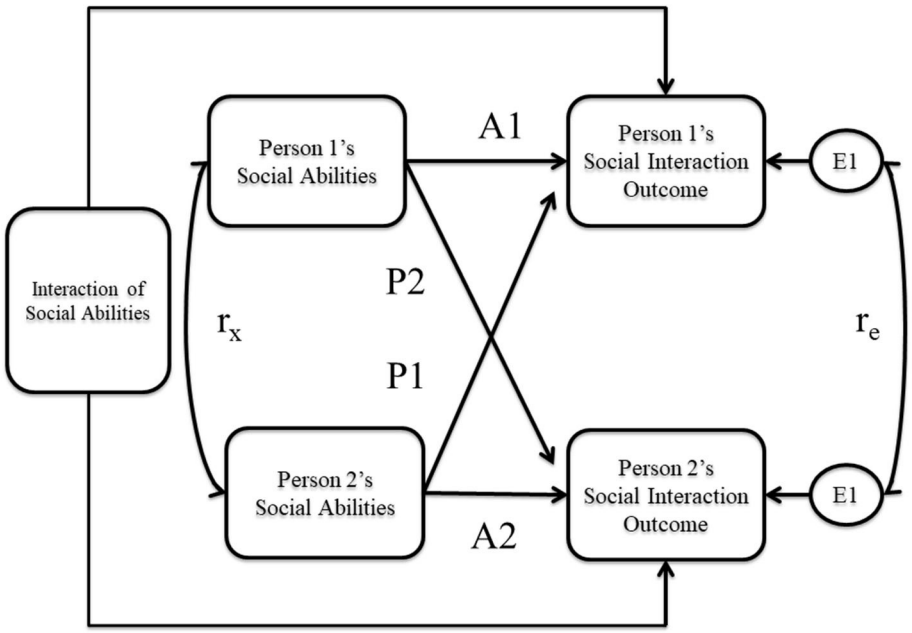

FIGURE 1 | Actor partner interdependence model (APIM) predicting social interaction outcomes with individual and partner social abilities. A-paths represent the actor effects and P-paths represent the partner effects. The interaction term represents the effect of the individual's social abilities on the individual's social interaction outcome depending on the partner's social abilities.

TABLE 3 | Scores on predictors for diagnostic and dyad groups.

	A-A dyads (n = 42)		NA-NA dyads (n = 40)		A-NA dyads (A left; NA right) (n = 42)				A overall (n = 66)		NA overall (n = 58)	
	M	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
Benton	43.35	4.26	47.35	3.64	43.14	3.95	46.00	3.43	43.28	4.12	46.93	3.60
TASIT	51.60	6.54	56.33	4.39	53.14	6.38	55.22	3.06	52.13	6.48	55.98	4.03
ER-40	34.08	2.49	34.73	2.72	33.52	2.73	34.56	2.28	33.89	2.57	34.67	2.57
Social Cog	−0.24	0.66	0.40	0.60	−0.24	0.70	0.21	0.47	−0.24	0.66	0.34	0.57
FMS	15.35	7.15	19.40	5.29	16.33	7.16	20.33	4.45	15.69	7.11	19.69	5.02
Overall_SS	5.71	1.00	6.35	0.63	5.67	0.96	6.65	0.56	5.57	1.04	6.44	0.62

M, Mean; SD, Standard Deviation; ER-40, Emotion Recognition test; FMS, Friendship Motivation Scale; SS, Social Skills; TASIT, The Awareness of Social Inference Test.

outcomes, and (b) if these effects are moderated by diagnosis and dyad type (see **Tables 7–15**). Each of the tables focuses on a different set of social ability predictors and social interaction outcome variables. Whereas, **Tables 7–9** includes APIM analyses with social evaluation measures as outcome variables, **Tables 10–15** include APIM analyses with first impression measures as outcome variables. Further, each table specifies the particular social ability variables being used as predictors (i.e., social cognitive, social skills, or social motivation).

For ease of presentation, we grouped the regression coefficients and standard errors for each set of predictors into different sections: (1) demographic variables, (2) diagnosis variables (i.e., the actor, partner, and actor-partner interaction effects for diagnostic status), (3) social ability variables (i.e., the actor, partner, and actor-partner interactions effects for the social ability variables), (4) moderation of the social ability variables by diagnosis variables (i.e., whether the actor, partner, and actor-partner interactions effects for the social ability variables depend upon the participants' or their partners' diagnostic status), and (5) moderation of the social ability variables by diagnostic combination or dyad type (i.e., whether the actor, partner, and actor-partner interactions effects for the social ability variables depend upon the diagnostic composition of the dyad). The regression coefficients and standard errors reflect the estimates from models wherein all predictors are included. Given the presence of multiple interaction terms, tolerance values (indices for multicollinearity) were rather low for the terms involving the social ability variables (mean values ranged from 0.26 for the social skill variables to 0.41 for the social motivation variables), moderation of the social ability variables

TABLE 4 | Correlations between predictors.

	Benton	TASIT	ER-40	FMS	Overall SS
Benton	1	0.017	0.361**	−0.193	0.035
TASIT	0.272*	1	0.221	−0.24	0.122
ER-40	0.121	0.308*	1	−0.009	0.196
FMS	0.129	0.166	0.278*	1	−0.037
Overall SS	0.06	0.335**	−0.137	−0.025	1

*NA correlations are above diagonal and A below it. Predictors are actor social abilities. FMS, Friendship Motivation Scale; ER-40, Emotion Recognition task; SS, Social Skill; TASIT, The Awareness of Social Inference test. *p< 0.05, **p < 0.01.*

by the diagnosis variables (mean values ranged from 0.25 for the social skill variables to 0.43 for the social motivation variables), and moderation of the social ability variables by diagnostic combination (mean values ranged from 0.26 for the social skill variables to 0.41 for the social motivation variables). Given our focus on the interaction terms, these tolerance values do not pose a problem (see McClelland et al., 2017).

Effects of Social Cognition on Interaction Outcomes

There were no significant actor, partner, or actor-partner interaction effects of social-cognition on any of the outcomes (see **Tables 7**, **10**, **13**). Nevertheless, we found a significant three-way interaction between actor diagnosis, partner diagnosis, and actor social cognition on interaction quality. To break this down, we inspected the simple two-way interactions between partner diagnosis and actor social cognition for autistic and NA participants. These analyses revealed an effect for NA actors ($b = 1.13$, SE $= 0.43$, $p = 0.01$) but not autistic actors ($b = -0.26$, SE $= 0.19$, $p = 0.18$). Within A-NA dyads, NA actors with higher social cognitive performance rated the interaction quality higher ($b = 1.80$, SE $= 0.72$, $p = 0.014$). However, this pattern was not observed for NA actors within NA-NA dyads ($b = -0.45$, SE $= 0.43$, $p = 0.30$).

We also found a significant two-way interaction of actor diagnosis and partner social cognition on awkwardness scores. Autistic adults rated partners with higher social cognitive performance as more awkward than partners with lower social cognitive performance ($b = -0.65$, SE $= 0.27$, $p = 0.02$). This pattern was not observed for NA adults ($b = 0.29$, SE $= 0.19$, $p = 0.13$). However, this interaction was subsumed by a three-way interaction of actor and partner diagnosis and partner social cognitive ability. To break this down, we first inspected the simple two-way interactions of partner diagnosis and partner social cognition for autistic actors and for NA actors. The two-way interaction was significant for autistic actors ($b = 0.61$, SE $= 0.27$, $p = 0.03$) but not NA actors ($b = -0.31$, SE $= 0.19$, $p = 0.10$). Further breaking down the two-way interaction for autistic actors revealed an effect of partner social cognition on autistic actors' awkwardness ratings in mixed dyads but not dyads of the same diagnosis. Specifically, whereas autistic actors rated their NA partners as more awkward when their partner had higher social cognitive performance ($b = -1.26$, SE $= 0.52$, $p = 0.02$), this effect was not seen for autistic actors in A-A dyads ($b = -0.03$, SE $= 0.15$, $p = 0.81$). No other moderating effects were observed.

TABLE 5 | Correlations between actor social abilities with actor outcomes.

NA	Benton	TASIT	ER-40	FMS	Overall social skill
Interaction quality	−0.068	0.058	−0.014	0.141	−0.048
Closeness	0.09	−0.152	−0.185	0.038	−0.174
IPC warmth	−0.159	0.096	−0.299*	0.004	−0.106
IPC dominance	−0.141	−0.313*	−0.181	−0.038	0.084
Awkward_R	0.039	−0.085	0.041	−0.034	−0.064
Attractive	−0.108	0.177	0.118	0.263*	0.062
Trustworthy	−0.214	0.300*	−0.147	−0.077	0.008
Aggressive/Dominant	−0.18	−0.188	0.053	0.008	−0.245
Likable	−0.087	0.099	−0.116	0.178	−0.001
Smart	−0.031	0.232	0.027	0.118	0.211
Live near	−0.108	0.235	−0.133	0.078	0.046
Hangout	0.208	0.178	−0.064	0.153	−0.039
Sit near	−0.05	0.138	−0.018	0.177	−0.094
Conversation	0.256	0.202	0.071	0.166	0.232
Behavioral Intent	0.094	0.278*	−0.062	0.209	0.041
A					
Interaction quality	0.143	0.013	−0.03	0.089	−0.135
Closeness	−0.093	−0.254*	−0.131	0.006	0.012
IPC warmth	0.019	−0.053	−0.166	0.409**	−0.021
IPC dominance	0.062	−0.316*	−0.153	−0.168	0.014
Awkward_R	−0.16	−0.105	−0.096	−0.026	−0.126
Attractive	0.044	0.024	0.018	−0.121	0.056
Trustworthy	−0.128	−0.051	−0.173	0.049	0.15
Aggressive/Dominant	0.042	0.132	0.042	−0.243*	0.22
Likable	−0.087	−0.190	−0.052	0.081	−0.006
Smart	−0.059	−0.015	0.135	0.290*	−0.008
Live near	0.087	0.002	0.250*	0.18	0.053
Hangout	−0.149	−0.305*	−0.038	0.19	−0.157
Sit near	0.111	−0.004	0.131	0.232	−0.049
Conversation	0.024	−0.162	−0.052	0.448**	0.037
Behavioral intent	0.039	−0.150	0.127	0.366**	−0.035

*Outcomes are actor ratings of the partner and interaction. Awkward was reverse scored. ER-40, Emotion Recognition task; FMS, Friendship Motivation Scale; IPC, Interpersonal Circumplex; TASIT, The Awareness of Social Inference test. *p < 0.05, **p < 0.01.*

Exploratory Analyses: Effects of Individual Social Cognitive Domains on Interaction Outcomes

In addition to examining effects of the overall social cognition composite, we explored the effects of performance on each individual social cognitive task (i.e., Benton, ER40, TASIT). There were significant two-way interactions of actor and partner diagnosis with partner emotion recognition abilities (i.e., ER-40 scores) for trustworthiness ratings. Autistic actors trusted their partners more when their partners had higher levels of emotion recognition ability ($b = 0.10$, SE $= 0.03$, $p = 0.003$). This effect was not significant for NA actors ($b = -0.03$, SE $= 0.03$, $p = 0.296$). Additionally, participants rated NA participants with stronger emotion recognition abilities as more trustworthy ($b =$

TABLE 6 | Correlations between actor predictors and partner outcomes.

NA	Benton	TASIT	ER-40	FMS	Overall social skill
Interaction quality	0.119	0.137	0.118	−0.053	0.123
Closeness	0.082	0.071	0.028	−0.042	0.109
IPC warmth	0.297*	0.132	0.193	0.109	0.256
IPC dominance	0.136	0.162	0.095	0.031	0.229
Awkward_R	0.206	0.137	0.106	−0.091	0.233
Attractive	−0.144	−0.114	0.067	0.225	0.115
Trustworthy	0.078	−0.178	0.220	0.274	−0.095
Aggressive/Dominant	−0.061	0.22	−0.013	0.099	−0.017
Likable	−0.120	0.105	0.027	0.041	0.096
Smart	−0.020	−0.03	0.057	0.24	−0.064
Live near	−0.053	−0.035	0.009	0.163	0.043
Hangout	0.079	0.145	−0.102	−0.051	0.129
Sit near	0.035	0.068	0.150	−0.071	−0.220
Conversation	0.204	−0.153	0.013	−0.09	−0.005
Behavioral Intent	0.073	0.026	0.025	0.001	−0.017
A					
Interaction quality	−0.013	−0.004	0.010	−0.063	0.260*
Closeness	0.188	0.144	0.192	0.054	0.073
IPC warmth	0.006	0.036	0.162	−0.086	0.221
IPC dominance	0.04	−0.185	−0.267*	−0.093	0.060
Awkward_R	0.041	0.094	−0.262*	−0.029	0.328*
Attractive	−0.107	0.101	−0.045	−0.2	0.224
Trustworthy	−0.022	−0.184	−0.147	0.095	0.206
Aggressive/Dominant	0.221	−0.067	−0.201	0.027	0.096
Likable	0.074	0.059	0.150	0.026	0.059
Smart	0.054	0.348**	0.192	0.149	0.265*
Live near	−0.029	0.063	0.142	0.168	−0.147
Hangout	0.22	0.22	0.023	0.058	0.149
Sit near	−0.081	0.033	0.070	0.189	−0.045
Conversation	0.041	−0.047	−0.071	0.135	0.227
Behavioral Intent	0.047	0.109	0.079	0.234	0.052

*Outcomes are partner ratings of the actor and interaction. Awkward was reverse scored. ER-40, Emotion Recognition task; FMS, Friendship Motivation Scale; IPC, Interpersonal Circumplex; TASIT, The Awareness of Social Inference test. *p < 0.05, **p < 0.01.*

0.09, SE = 0.03, $p = 0.006$), but this effect was not significant for autistic partners with differing levels of emotion recognition ability ($b = -0.04$, SE = 0.03, $p = 0.15$).

There were also significant two-way interactions for likeability. Autistic actors liked partners more when their partners had higher emotion recognition abilities ($b = 0.09$, SE = 0.03, $p = 0.009$), but this effect was not significant for NA actors ($b = -0.03$, SE = 0.03, $p = 0.31$). The interaction of partner diagnosis with facial recognition scores (i.e., Benton) was also significant ($p = 0.002$). Following up this interaction with simple slopes revealed that the effect of Benton scores on likeability ratings did not significantly differ from zero for both autistic and NA partners, but the pattern of effects suggests that participants rated higher likeability for NA partners who had lower facial recognition scores (NA partner: $b = -0.04$, SE = 0.02, $p = 0.08$) and for autistic adults who had higher facial recognition scores (A partner: $b = 0.03$, SE = 0.02, $p = 0.08$).

Effects of Social Skills on Interaction Outcomes

There was a significant effect of the partner's composite social skills rating on awkwardness evaluations ($p < 0.001$), such that partners who were higher on observed social skills were rated as less awkward. No other actor, partner, or actor-partner interaction effects involving social skills were significant, and there was no evidence that diagnostic status or dyad type moderated any of these effects (see **Tables 8**, **11**, **14**).

Effects of Social Motivation on Interaction Outcomes

There were no significant actor, partner, or actor-partner interactions for the social motivation variables on any of the social interaction outcome variables (see **Tables 9**, **12**, **15**). However, there was a significant three-way interaction of actor and partner diagnoses with actor motivation scores on trustworthiness ratings ($p = 0.007$). To break this down, we examined the simple two-way interactions between partner diagnosis and actor social motivation for autistic and NA actors. There was a significant interaction of partner diagnosis with actor social motivation for NA actors ($b = -0.03$, SE = 0.02, $p = 0.03$). NA actors with more social motivation rated their autistic partners as less trustworthy ($b = -0.06$, SE = 0.03, $p = 0.04$), but this did not extend to NA partners ($b = 0.01$, SE = 0.01, $p = 0.45$). Moreover, the interaction of partner diagnosis and social motivation was marginally significant for autistic actors ($b = 0.02$, SE = 0.01, $p = 0.06$). Breaking this two-way interaction down further revealed that autistic actors with more social motivation rated other autistic adults as marginally more trustworthy ($b = 0.02$, SE = 0.01, $p = 0.06$), but this effect did not extend to NA partners ($b = -0.02$, SE = 0.02, $p = 0.26$).

DISCUSSION

In a previous study using this sample (Morrison et al., 2020), autistic adults were evaluated less favorably by unfamiliar partners following a "get to know you" conversation, and NA participants were less interested than autistic participants in interacting with them again in the future. In the current study, these autistic adults performed lower on a composite of social cognitive measures, were rated as less normative on social skills, and endorsed fewer normative indicators of social motivation compared to NA adults. All of these findings align with previous research (Chevallier et al., 2012a; Morrison et al., 2017, 2019a; Sasson et al., 2017; DeBrabander et al., 2019), but contrary to expectation, only minimal links were found between autistic adults' performance on the three social ability domains and their social interaction outcomes. In some cases, it was the social abilities of NA adults, not those of autistic adults, that were most predictive of outcomes, and this was particularly the case when they were interacting with autistic people. NA social cognition, for instance, predicted some of their interaction outcomes (e.g., awkwardness, interaction quality) with autistic but not NA partners. Collectively, findings suggest that standalone measures of autistic social abilities are not particularly predictive of

TABLE 7 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social cognition variables on the social evaluation outcomes of closeness, interaction quality, warmth, and dominance.

	Social evaluation outcomes							
Predictors	**Closeness**		**Interaction quality**		**IPC warmth**		**IPC dominance**	
Social-cognition predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	2.92	0.23	5.70	0.18	0.03	0.17	−0.03	0.20
Demographic variables								
Actor WRAT	−0.02	0.01	−0.01	0.01	0.01	0.01	−0.01	0.01
Actor race—AfricanAmerican	−0.01	0.42	0.11	0.34	0.37	0.31	0.06	0.36
Actor race—Asian	0.18	0.46	−0.51	0.37	−0.35	0.34	−0.33	0.39
Actor race—Other	0.05	0.34	0.52	0.29	0.09	0.27	0.32	0.31
Actor age	0.05	0.03	0.06*	0.02	0.06*	0.02	0.02	0.03
Diagnosis variables								
Actor diagnosis	0.24	0.15	−0.03	0.13	−0.20	0.12	−0.16	0.14
Partner diagnosis	−0.04	0.14	−0.12	0.12	−0.14	0.11	−0.17	0.13
Actor*Partner diagnosis	0.19	0.16	0.12	0.13	0.03	0.12	0.03	0.14
Social cognition variables								
Actor SC	−0.13	0.26	0.43	0.22	0.06	0.21	−0.21	0.24
Partner SC	0.38	0.26	0.13	0.22	0.40	0.21	−0.15	0.24
Actor*Partner SC	−0.30	0.39	0.14	0.32	0.09	0.29	0.31	0.34
Moderation of social cognition variables by diagnosis variables								
Actor diagnosis*Actor SC	−0.12	0.26	−0.25	0.22	−0.10	0.20	−0.05	0.24
Actor diagnosis*Partner SC	0.06	0.27	−0.08	0.23	0.15	0.21	−0.14	0.25
Actor diagnosis*(Actor*Partner SC)	−0.02	0.32	−0.30	0.28	−0.03	0.26	0.09	0.31
Partner diagnosis*Actor SC	−0.04	0.28	0.43	0.23	0.18	0.22	0.18	0.26
Partner diagnosis*Partner SC	−0.03	0.26	−0.16	0.22	−0.31	0.20	−0.14	0.24
Partner diagnosis*(Actor*Partner SC)	−0.02	0.32	0.53	0.28	0.41	0.26	0.10	0.31
Moderation of social cognition variables by dyad type								
(Actor*Partner diagnosis)*Actor SC	−0.22	0.28	−0.69**	0.24	−0.34	0.22	−0.40	0.26
(Actor*Partner diagnosis)* Partner SC	−0.15	0.28	−0.17	0.23	−0.27	0.22	0.05	0.25
(Actor*Partner diagnosis)* (Actor*Partner SC)	−0.14	0.38	−0.19	0.31	−0.09	0.29	0.04	0.33

*IPC, Interpersonal Circumplex; WRAT-3, Wide Range Achievement Test – 3; SC, Social Cognition. All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. *$p < 0.05$, **$p < 0.01$.*

their poorer interaction outcomes with NA partners. Rather, more consistent with relational accounts of autistic sociability (Milton, 2012; Bottema-Beutel, 2017; Bolis et al., 2018; Redcay and Schilbach, 2019), the dyadic combination of social abilities between diagnostic groups was more predictive of how autistic and NA adults evaluated (and were evaluated by) their partners.

Across the three social abilities assessed here, only normative social skill demonstrated any unidirectional predictive value on interaction outcomes. Most notably, those who were coded as less normative in their overall social skill were evaluated as more awkward. It may be the case that the overall social skill rating used here (Pinkham and Penn, 2006) is driven in part by the coder's perception of the person's awkwardness, which tended to align with participant evaluations of awkwardness within the dyads. This interpretation may explain why the social skill measure was associated with awkwardness ratings but not other evaluated traits: "awkwardness" may be consistent with an individual's judgment of another person's social skill, with lower ratings signifying a deviation from normative social expression and behavior. Perhaps not coincidently, NA raters in previous studies have tended to discriminate autistic and NA participants more on awkwardness than any other trait judgment (Grossman, 2015; Sasson et al., 2017; Sasson and Morrison, 2019), with awkwardness ratings highly associated with a reluctance among NA adults to pursue subsequent social interaction.

Autistic raters in this study also judged autistic people high on awkwardness, but unlike NA raters, this judgment was not associated with reduced social interest (Morrison et al., 2020). What underlies this dissociation remains unclear. Future research may seek to isolate the specific characteristics and cues driving higher scores of awkwardness and assess whether they may be interpreted and valued differently by autistic and NA people. For instance, recent findings suggest that autistic people may seek out interaction with those who present and communicate atypically (Granieri et al., 2020), as these differences—ones often described as "awkward"

TABLE 8 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social skills variables on the social evaluation outcomes of closeness, interaction quality, warmth, and dominance.

	Social evaluation outcomes							
Predictors	**Closeness**		**Interaction quality**		**IPC warmth**		**IPC dominance**	
Social skills predictors	**b**	**SE**	**b**	**SE**	**b**	**SE**	**b**	**SE**
Intercept	2.90	0.24	5.86	0.21	−0.04	0.20	−0.23	0.22
Demographic variables								
Actor WRAT	−0.02	0.01	−0.01	0.01	0.00	0.01	−0.01	0.01
Actor race – AfricanAmerican	0.15	0.39	0.21	0.34	0.23	0.33	−0.28	0.36
Actor race – Asian	0.01	0.48	−0.49	0.42	−0.09	0.40	−0.05	0.44
Actor race – Other	0.05	0.33	0.53	0.29	0.04	0.28	0.42	0.32
Actor age	0.06*	0.03	0.04	0.03	0.06*	0.02	0.00	0.03
Diagnosis variables								
Actor diagnosis	0.31	0.16	−0.10	0.15	−0.27	0.14	0.06	0.17
Partner diagnosis	−0.02	0.16	0.09	0.15	−0.00	0.14	−0.09	0.16
Actor*Partner diagnosis	0.12	0.21	−0.00	0.19	0.12	0.18	0.19	0.20
Social skills variables								
Actor SS	−0.06	0.19	−0.18	0.17	−0.21	0.16	0.20	0.18
Partner SS	0.28	0.19	0.38*	0.17	0.38*	0.16	0.29	0.19
Actor*Partner SS	−0.34	0.23	−0.29	0.20	0.15	0.19	0.17	0.21
Moderation of social skills variables by diagnosis variables								
Actor diagnosis*Actor SS	0.08	0.22	0.17	0.19	0.02	0.18	−0.18	0.20
Actor diagnosis*Partner SS	−0.25	0.21	−0.22	0.19	0.07	0.18	−0.13	0.20
Actor diagnosis*(Actor*Partner SS)	0.40*	0.19	0.16	0.18	0.12	0.17	0.10	0.19
Partner diagnosis*Actor SS	−0.09	0.21	−0.13	0.18	0.17	0.17	0.42*	0.19
Partner diagnosis*Partner SS	−0.24	0.22	0.03	0.20	−0.27	0.19	−0.23	0.21
Partner diagnosis*(Actor*Partner SS)	0.18	0.19	0.00	0.18	−0.12	0.17	0.00	0.19
Moderation of social skills variables by dyad type								
(Actor*Partner diagnosis)*Actor SS	0.14	0.19	0.02	0.17	−0.03	0.16	−0.29	0.18
(Actor*Partner diagnosis)*Partner SS	0.34	0.19	−0.06	0.17	−0.13	0.16	0.02	0.19
(Actor*Partner diagnosis)*(Actor*Partner SS)	0.01	0.24	0.23	0.21	−0.06	0.20	−0.07	0.22

*IPC, Interpersonal Circumplex; WRAT-3, Wide Range Achievement Test – 3; SS, Social Skills. All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. *$p < 0.05$.*

—may cohere with their social preferences and facilitate better interpersonal communication and connection. Similarly, Heasman and Gillespie (2019) found that, contrary to normative expectations, misunderstanding, and misinterpretation among autistic adults did not invariably lead to deterioration of the interaction. Viewed through a conventional social lens, such disruptions may be perceived as awkward or seen as evidence of social disjunction, but these instances may be experienced differently by autistic adults. Perceptions of "awkwardness" therefore may reflect just one of many differences in social expectations and experiences between autistic and NA people.

Aside from ratings of awkwardness, normative social skill did not predict any trait evaluations or interaction outcomes for either the individual or their partner, was no more predictive of outcomes for autistic compared to NA adults, and did not vary across different dyad combinations. It may be the case that broader social judgments within a "get to know you" conversation depend less upon observable social skill and more upon other characteristics and considerations. For example, ratings of traits such as attractiveness may be influenced more by physical attributes rather than social behaviors, and judgments of likeability, trustworthiness, warmth, and interaction quality may be more related to conversational content, personal disclosure, and interpersonal alignment. Alternatively, or perhaps complementarily, conceptualizing social skill as an objective metric in which individuals can be quantitatively rank ordered and a single standard applied to all populations may be unhelpful for predicting complex social relationship dynamics, particularly between neurologically diverse people (Heerey, 2015; Bottema-Beutel, 2017; Milton, 2017). What constitutes good "social skill" may vary across groups and individuals, and a single holistic social skill rating may simply be inadequate for capturing and summarizing social skill across an entire dynamic and emergent interaction. Indeed, work examining interpersonal warmth and dominance suggests that moment to moment behaviors rather than overall summaries

TABLE 9 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social motivation variables on the social evaluation outcomes of closeness, interaction quality, warmth, and dominance.

	Social evaluation outcomes							
Predictors	**Closeness**		**Interaction quality**		**IPC warmth**		**IPC dominance**	
Social motivation predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	2.87	0.24	5.78	0.21	0.11	0.16	0.04	0.22
Demographic variables								
Actor WRAT	−0.03*	0.01	−0.01	0.01	0.01	0.01	−0.02*	0.01
Actor race – AfricanAmerican	0.32	0.40	0.08	0.34	0.28	0.27	−0.04	0.36
Actor race – Asian	0.05	0.45	−0.43	0.39	−0.21	0.30	−0.23	0.40
Actor race – Other	−0.18	0.33	0.67*	0.30	0.17	0.25	0.39	0.32
Actor age	0.05*	0.03	0.04	0.02	0.05*	0.02	−0.00	0.03
Diagnosis variables								
Actor diagnosis	0.35**	0.12	0.16	0.11	−0.06	0.10	−0.00	0.13
Partner diagnosis	−0.12	0.12	−0.16	0.11	−0.16	0.10	−0.09	0.12
Actor*Partner diagnosis	0.23	0.14	0.13	0.12	0.10	0.10	−0.03	0.13
Social motivation variables								
Actor SM	0.02	0.02	0.03	0.02	0.02	0.02	−0.00	0.02
Partner SM	0.01	0.02	−0.02	0.02	−0.00	0.02	−0.02	0.02
Actor*Partner SM	0.01	0.00	0.00	0.00	−0.00	0.00	0.00	0.00
Moderation of social motivation variables by diagnosis variables								
Actor diagnosis*Actor SM	−0.01	0.02	−0.02	0.02	0.00	0.02	0.01	0.02
Actor diagnosis*Partner SM	−0.00	0.02	0.01	0.02	0.00	0.02	−0.01	0.02
Actor diagnosis*(Actor*Partner SM)	−0.00	0.00	0.00	0.00	0.00	0.00	−0.00	0.00
Partner diagnosis*Actor SM	0.02	0.02	0.00	0.02	0.04*	0.02	−0.01	0.02
Partner diagnosis*Partner SM	0.02	0.02	0.00	0.02	−0.02	0.02	0.00	0.02
Partner diagnosis *(Actor*Partner SM)	0.00	0.00	0.00	0.00	0.00	0.00	0.01	0.00
Moderation of social motivation variables by dyad type								
(Actor*Partner diagnosis)*Actor SM	−0.02	0.02	−0.00	0.02	0.02	0.02	0.00	0.02
(Actor*Partner diagnosis)* Partner SM	−0.00	0.02	0.01	0.02	−0.01	0.02	0.04	0.02
(Actor*Partner diagnosis)* (Actor*Partner SM)	−0.00	0.00	−0.00	0.00	−0.00	0.00	−0.00	0.00

*IPC, Interpersonal Circumplex; WRAT-3, Wide Range Achievement Test – 3; SM, Social Motivation; All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. *$p < 0.05$, **$p < 0.01$.*

are better predictors of interaction outcomes (Markey et al., 2010; Stevanovic et al., 2017).

Autistic adults' social cognitive performance was also not particularly predictive of their interaction outcomes. In fact, within mixed dyads, it was the social cognitive performance of NA adults, not autistic adults, that generated most of the effects. For instance, better social cognitive performance among NA adults was associated with rating conversations with autistic partners as higher in quality. One possible interpretation is that social cognitive ability among NA adults may facilitate better perception of social cues from their autistic partners and mitigate the difficulties NA people often have inferring autistic mental states (Edey et al., 2016; Sheppard et al., 2016; Gernsbacher et al., 2017). This also suggests that interaction quality between autistic and NA adults may improve by increasing social cognitive ability among NA people—perhaps more so than among autistic people given that no corresponding effect was found for autistic participants. Indeed, some emerging evidence indicates that NA observers who are better able to infer autistic mental states (Alkhaldi et al., 2019) and have greater understanding about autism (Sasson and Morrison, 2019) evaluate autistic people more favorably, suggesting that social experiences of autistic people within NA environments may improve with greater NA understanding about autism. Less provocatively, higher social cognitive performance among NA adults in this study may have been a proxy for other characteristics associated with more enjoyable conversational experiences with autistic partners, like higher social engagement, attentiveness, and desire for connection. Regardless of interpretation, however, this finding of NA social cognition predicting outcomes with autistic partners was not hypothesized and should therefore be interpreted cautiously until replicated.

Although NA adults with higher social cognitive performance rated conversations with autistic partners as higher in quality, autistic participants did not share this assessment and instead actually perceived NA adults who scored better on social

TABLE 10 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social cognition variables on the first impression variables of behavioral intent, awkwardness (reversed), attractiveness, and trustworthiness.

Predictors	First-impression outcome variables							
	Behavioral intent		Awkwardness (reverse scored)		Attractiveness		Trustworthiness	
Social-cognition predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	3.09	0.09	3.03	0.12	2.51	0.16	3.42	0.09
Demographic variables								
Actor WRAT	0.00	0.00	−0.01	0.01	0.01	0.01	0.00	0.01
Actor race – AfricanAmerican	0.13	0.17	0.32	0.22	0.11	0.30	0.04	0.17
Actor race – Asian	−0.36	0.18	−0.24	0.24	−0.04	0.32	0.15	0.18
Actor race – Other	0.27	0.14	0.03	0.21	−0.00	0.24	0.05	0.15
Actor age	0.03*	0.01	0.02	0.02	0.03	0.02	−0.00	0.01
Diagnosis variables								
Actor diagnosis	−0.04	0.06	0.06	0.09	0.08	0.11	−0.08	0.06
Partner diagnosis	−0.03	0.06	−0.28**	0.09	−0.16	0.10	−0.07	0.06
Actor*Partner diagnosis	0.14*	0.07	−0.03	0.09	−0.08	0.12	0.06	0.07
Social cognition variables								
Actor SC	0.08	0.11	0.15	0.16	0.32	0.19	−0.16	0.11
Partner SC	0.02	0.11	−0.18	0.16	0.02	0.19	−0.06	0.11
Actor*Partner SC	0.15	0.16	−0.01	0.21	0.08	0.28	0.06	0.16
Moderation of social cognition variables by diagnosis variables								
Actor diagnosis*Actor SC	−0.09	0.11	−0.32	0.16	−0.25	0.19	0.08	0.11
Actor diagnosis*Partner SC	0.11	0.11	−0.47**	0.17	−0.13	0.20	0.13	0.12
Actor diagnosis*(Actor*Partner SC)	−0.10	0.14	−0.03	0.21	−0.01	0.23	−0.18	0.14
Partner diagnosis*Actor SC	0.17	0.12	0.29	0.17	0.12	0.20	0.06	0.12
Partner diagnosis*Partner SC	−0.03	0.11	0.15	0.16	−0.03	0.19	−0.14	0.11
Partner diagnosis*(Actor*Partner SC)	0.28*	0.14	0.50*	0.21	0.05	0.23	0.08	0.14
Moderation of social cognition variables by dyad type								
(Actor*Partner diagnosis)*Actor SC	−0.09	0.12	−0.10	0.17	−0.26	0.20	−0.00	0.12
(Actor*Partner diagnosis)*Partner SC	−0.02	0.12	0.46**	0.17	0.08	0.20	−0.09	0.12
(Actor*Partner diagnosis)*(Actor*Partner SC)	−0.07	0.15	0.14	0.20	−0.34	0.27	−0.06	0.15

*WRAT−3, Wide Range Achievement Test – 3; SC, Social Cognition. All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. *$p < 0.05$, **$p < 0.01$.*

cognitive measures as more awkward than those who scored lower. At first blush, this finding appears counterintuitive and potentially spurious, but the strict alpha level reduces the likelihood that this is the case. What underlies this effect is unclear, but it may be the case that social cognitive ability among NA individuals manifests in social behaviors perceived as awkward or intrusive by autistic adults. Alternatively, as suggested previously, autistic individuals may interpret the term "awkward" differently than NA individuals, but other findings suggest that autistic adults did perceive "awkward" as a negative characteristic—their ratings of awkwardness were related to lower intentions to interact as well as with other less favorable trait evaluations. Importantly, however, these relationships were weaker than those found for NA adults.

Additionally, despite performing lower on several social cognitive tasks, autistic adults largely mirrored NA adults in forming less favorable evaluations of other autistic adults (Morrison et al., 2020). Thus, contrary to what might be expected based on their lower social cognitive performance, autistic adults appeared just as sensitive to social presentation differences among autistic adults and interpreted these differences similarly to their NA counterparts. Additionally, autistic adults rated partners who were more skilled in emotion recognition ability as more trustworthy and likable. This suggests that despite performing less well on standalone social cognitive tasks, interacting with someone skilled in these domains improved how autistic adults perceived their partner. Taken together, these findings suggest that the lower social cognitive performance demonstrated by autistic adults did not correspond in clear and predictable ways to their real-world social interaction outcomes. Isolated computerized assessments of social cognition such as those used here may not fully capture how these social abilities influence actual social interaction. This does not mean that these measures fail to capture social cognitive

TABLE 11 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social skills variables on the first impression variables of behavioral intent, awkwardness (reversed), attractiveness, and trustworthiness.

Predictors	First-impression outcome variables							
	Behavioral intent		Awkwardness (reverse scored)		Attractiveness		Trustworthiness	
Social skills predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	3.09	0.10	2.98	0.13	2.60	0.17	3.48	0.10
Demographic variables								
Actor WRAT	0.00	0.01	−0.01	0.01	0.01	0.01	0.01	0.01
Actor race – AfricanAmerican	0.20	0.16	−0.06	0.22	0.05	0.28	0.19	0.17
Actor race – Asian	−0.30	0.20	−0.21	0.27	−0.27	0.34	0.13	0.20
Actor race – Other	0.15	0.14	0.25	0.20	0.21	0.23	−0.12	0.15
Actor age	0.02	0.01	0.01	0.02	0.02	0.02	−0.01	0.01
Diagnosis variables								
Actor diagnosis	0.05	0.08	−0.08	0.13	−0.03	0.12	0.10	0.08
Partner diagnosis	−0.06	0.07	−0.18	0.12	−0.04	0.12	−0.14	0.08
Actor*Partner diagnosis	0.09	0.09	0.06	0.12	−0.25	0.15	−0.09	0.09
Social skills variables								
Actor SS	0.03	0.08	−0.08	0.13	0.00	0.13	0.04	0.09
Partner SS	0.01	0.08	0.38**	0.13	0.21	0.13	−0.07	0.09
Actor*Partner SS	−0.17	0.09	−0.04	0.13	−0.17	0.16	−0.07	0.10
Moderation of social skills variables by diagnosis variables								
Actor diagnosis*Actor SS	−0.02	0.09	−0.04	0.13	0.11	0.15	0.08	0.10
Actor diagnosis*Partner SS	−0.07	0.09	0.02	0.13	−0.29	0.15	−0.10	0.10
Actor diagnosis*(Actor*Partner SS)	−0.03	0.09	0.09	0.14	0.26	0.14	−0.08	0.09
Partner diagnosis*Actor SS	−0.02	0.09	0.03	0.13	−0.23	0.15	−0.04	0.09
Partner diagnosis*Partner SS	0.01	0.09	−0.09	0.14	0.22	0.16	0.11	0.10
Partner diagnosis *(Actor*Partner SS)	0.05	0.09	−0.00	0.14	−0.06	0.14	0.06	0.09
Moderation of social skills variables by dyad type								
(Actor*Partner diagnosis)*Actor SS	−0.01	0.08	−0.00	0.13	0.17	0.13	−0.10	0.09
(Actor*Partner diagnosis)*Partner SS	0.06	0.08	−0.03	0.13	−0.00	0.13	0.11	0.09
(Actor*Partner diagnosis)* (Actor*Partner SS)	0.05	0.10	−0.07	0.13	0.23	0.17	0.02	0.10

*WRAT-3, Wide Range Achievement Test – 3; SS, Social Skills. All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. **$p < 0.01$.*

differences; in fact, as in previous research (Morrison et al., 2019b), they differentiated autistic and NA participants and were somewhat predictive of NA outcomes. Nevertheless, current findings raise questions about the mechanistic link between reduced social cognitive performance by autistic adults on standalone tasks and their difficulties interacting with NA adults. Recognizing faces and emotions from static images may not translate in presumed ways to the much more complex nature of dynamic interaction, even within NA-NA interactions. Similarly, higher social cognitive performance by NA adults did not facilitate better mutual interaction quality or an increase in shared positive outcomes with autistic adults. Collectively, such findings are consistent with double empathy (Milton, 2012) and dialectical misattunement (Bolis et al., 2018) theories of social disconnection between autistic and NA people and suggest that traditional conceptualizations of social cognitive ability may not extend in anticipated ways to autistic-NA interactions.

For social motivation, moderated results suggested lower social motivation scores among autistic participants did not impact how they were evaluated in the conversation. Indeed, there was only one group effect of social motivation, such that NA adults high on social motivation trusted their autistic partners less. It may be the case that socially motivated NA adults strive but struggle to connect with their autistic partners and misinterpret autistic social differences as indicative of lower trustworthiness. If so, this process could have adverse consequences for the social experiences of autistic adults, whose differences in social expressivity could be misperceived in ways that reinforce reluctance of NA adults to interact with them. Such an interpretation, however, is currently speculative and worthy of verification in future study.

Taken together, results from this study challenge traditional thinking about the mechanisms of social interaction difficulties

TABLE 12 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social motivation variables on the first impression variables of behavioral intent, awkwardness (reversed), attractiveness, and trustworthiness.

	First-impression outcome variables							
Predictors	**Behavioral intent**		**Awkwardness (reverse scored)**		**Attractiveness**		**Trustworthiness**	
Social Motivation Predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	3.09	0.09	3.14	0.14	2.55	0.16	3.48	0.09
Demographic variables								
Actor WRAT	0.00	0.00	−0.01	0.01	0.01	0.01	0.01	0.00
Actor race – AfricanAmerican	0.17	0.15	−0.00	0.23	−0.11	0.26	0.03	0.15
Actor race – Asian	−0.31	0.16	−0.15	0.26	0.08	0.30	0.30	0.16
Actor race – Other	0.20	0.13	0.22	0.21	0.20	0.23	−0.04	0.14
Actor age	0.02	0.01	0.02	0.02	0.02	0.02	−0.00	0.01
Diagnosis variables								
Actor diagnosis	0.06	0.05	0.10	0.09	0.07	0.09	−0.02	0.06
Partner diagnosis	−0.03	0.05	−0.43**	0.09	−0.20*	0.08	0.02	0.05
Actor*Partner diagnosis	0.13*	0.05	−0.01	0.08	−0.03	0.09	−0.03	0.05
Social motivation variables								
Actor SM	0.02*	0.01	0.01	0.02	0.01	0.01	−0.01	0.01
Partner SM	0.00	0.01	−0.03	0.02	−0.01	0.01	0.01	0.01
Actor*Partner SM	0.00	0.00	−0.00	0.00	−0.01*	0.00	−0.00*	0.00
Moderation of social motivation variables by diagnosis variables								
Actor diagnosis*Actor SM	−0.00	0.01	−0.00	0.01	−0.03	0.02	0.01	0.01
Actor diagnosis*Partner SM	0.01	0.01	−0.01	0.01	−0.02	0.02	0.00	0.01
Actor diagnosis*(Actor*Partner SM)	0.00	0.00	−0.00	0.00	−0.00	0.00	0.00	0.00
Partner diagnosis*Actor SM	0.00	0.01	0.01	0.01	−0.01	0.02	−0.01	0.01
Partner diagnosis*Partner SM	−0.00	0.01	0.01	0.01	−0.01	0.02	−0.01	0.01
Partner diagnosis*(Actor*Partner SM)	0.00	0.00	0.00	0.00	0.00	0.00	0.00	0.00
Moderation of social motivation variables by dyad type								
(Actor*Partner diagnosis)*Actor SM	0.01	0.01	−0.00	0.02	0.00	0.01	0.03**	0.01
(Actor*Partner diagnosis)*Partner SM	−0.00	0.01	0.02	0.01	0.02	0.01	−0.00	0.01
(Actor*Partner diagnosis)* (Actor*Partner SM)	−0.00	0.00	0.00	0.00	0.00	0.00	0.00	0.00

*WRAT-3, Wide Range Achievement Test – 3; SM, Social Motivation. All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. *p < 0.05, **p < 0.01.*

for autistic adults. Typically, because on average autistic adults perform lower than NA controls on traditional social cognitive tasks (Morrison et al., 2019b), deviate in their social behavior and presentation from prototypical social skills (Morrison et al., 2017), and often report lower or different social motivation (Chevallier et al., 2012a), psychosocial treatments often seek to train autistic people to be more normative in these areas with the hope doing so will translate to greater social interaction success in the real-world (Bishop-Fitzpatrick et al., 2014; Kern Koegel et al., 2016). However, this result does not regularly occur in practice (Gates et al., 2017; Bottema-Beutel et al., 2018). Recent empirical work has shown that social cognitive performance and social skill among autistic adults on standardized measures demonstrate only a small correspondence to their functional outcomes beyond other factors (Sasson et al., 2020), and some autistic people can exhibit normative social skill despite lower theory of mind performance through cognitive compensation (Livingston et al., 2019). Indeed, among autistic adults without intellectual disability, general cognition is far more predictive of social skill than social cognition (Sasson et al., 2020), and performance on explicit social cognitive measures like the ones used here may be less predictive of social communication and interaction behavior in autism than implicit social cognitive performance (Keifer et al., 2020). Taken at face value, results from this study suggest that social cognition, social skill, and social motivation may not be useful treatment targets for improving autistic adults' initial social interactions with NA people. Alternatively, they may still influence real-life social outcomes in autism, but each were either poorly measured in the current study or done so in a way that has limited application to interaction outcomes. From this perspective, the field may improve from the development of more real-world assessments of social cognitive, social motivational, and social skills abilities, rather than continuing to rely solely on paper and pencil and computerized tasks.

TABLE 13 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social cognition variables on the first impression variables of aggressiveness/dominance, smartness, and liking.

	First-impression outcome variables					
Predictors	**Aggressiveness/Dominance**		**Smart**		**Liking**	
Social-cognition predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	1.74	0.10	3.46	0.14	3.35	0.09
Demographic variables						
Actor WRAT	−0.00	0.01	−0.01	0.01	0.00	0.01
Actor race – AfricanAmerican	0.05	0.18	−0.30	0.25	0.16	0.15
Actor race – Asian	0.08	0.20	0.25	0.27	−0.37*	0.17
Actor race – Other	0.05	0.18	0.26	0.22	0.30*	0.15
Actor age	0.01	0.02	0.06**	0.02	0.02	0.01
Diagnosis variables						
Actor diagnosis	0.16	0.09	−0.14	0.09	−0.06	0.07
Partner diagnosis	0.06	0.08	−0.04	0.09	0.01	0.07
Actor*Partner diagnosis	−0.00	0.07	0.21*	0.10	0.01	0.06
Social cognition variables						
Actor SC	0.13	0.15	0.05	0.17	0.08	0.12
Partner SC	0.07	0.15	−0.02	0.17	0.02	0.12
Actor*Partner SC	0.01	0.17	−0.00	0.23	−0.01	0.15
Moderation of social cognition variables by diagnosis variables						
Actor diagnosis*Actor SC	−0.13	0.15	0.06	0.16	−0.17	0.12
Actor diagnosis*Partner SC	−0.08	0.15	0.07	0.17	0.04	0.13
Actor diagnosis *(Actor*Partner SC)	0.10	0.20	−0.29	0.21	−0.03	0.17
Partner diagnosis*Actor SC	−0.01	0.16	0.26	0.18	0.18	0.13
Partner diagnosis*Partner SC	−0.18	0.15	0.25	0.16	0.01	0.12
Partner diagnosis*(Actor*Partner SC)	−0.02	0.20	0.20	0.21	0.33	0.17
Moderation of social cognition variables by dyad type						
(Actor*Partner diagnosis)* Actor SC	0.06	0.16	−0.25	0.18	−0.08	0.13
(Actor*Partner diagnosis)*Partner SC	0.10	0.15	−0.01	0.17	0.12	0.13
(Actor*Partner diagnosis)*(Actor*Partner SC)	−0.44*	0.17	0.14	0.23	−0.18	0.14

*WRAT−3, Wide Range Achievement Test – 3; SC, Social Cognition. All continuous variables were grand–mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. *$p < 0.05$, **$p < 0.01$.*

The field may also benefit from exploring how other abilities and behaviors of autistic adults may be predictive of how they are evaluated. Some recent work has argued that much remains unknown about social interaction in autism (Bottema-Beutel, 2017; Bottema-Beutel et al., 2018) and has suggested applying new theoretical frameworks for understanding autistic social interaction (Bottema-Beutel, 2017). For example, (Bottema-Beutel, 2017) contends that research on social abilities should be examined using sociolinguistic approaches (e.g., conversation analysis) which not only takes the individual's context into account, but also allows for more dynamic assessment of how a person interacts with his or her environment. Additionally, this kind of approach allows for the examination of environmental and societal influences such as stigma that may play a role in how social disability develops and manifests, as well as determining the efficacy of current interventions for treating social disability (Bottema-Beutel et al., 2018). Moreover, given the heterogeneity of autism, a more person-centered approach may better approximate understanding of social difficulties than the group-level assessments and analyses used in this study and most prior work.

This is particularly important because the group-level dyadic analyses pursued here may have been under-powered to detect some effects. The sample size was determined based upon medium to large effects reported in prior interaction studies, but these may have been artificially inflated because of their smaller sample sizes (Usher et al., 2018) or because they examined different populations like the Broad Autism Phenotype (Faso et al., 2016). As a result, the effects here may have been smaller than the medium or large effects that were anticipated, and thus may not have been detectable with the current sample size of 55 dyads. Relatedly, null effects from this study should not be treated as definitive, as some may have reached statistical significance with increased power. Another limitation of the current study is that it used only a few of the social cognitive, social motivational, and social skills assessments that exist,

TABLE 14 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social skills variables on the first impression variables of aggressiveness/dominance, smartness, and liking.

	First-impression outcome variables					
Predictors	**Aggressiveness/Dominance**		**Smart**		**Liking**	
Social skills predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	1.79	0.11	3.30	0.15	3.35	0.10
Demographic variables						
Actor WRAT	−0.01	0.01	−0.00	0.01	0.00	0.01
Actor race – AfricanAmerican	0.00	0.18	−0.22	0.24	0.13	0.17
Actor race – Asian	−0.02	0.22	0.28	0.30	−0.35	0.21
Actor race – Other	0.04	0.17	0.28	0.21	0.28	0.16
Actor age	0.02	0.02	0.03	0.02	0.02	0.01
Diagnosis variables						
Actor diagnosis	0.01	0.12	0.13	0.11	−0.04	0.10
Partner diagnosis	0.07	0.12	−0.13	0.11	−0.01	0.09
Actor*Partner diagnosis	−0.02	0.10	0.21	0.13	−0.06	0.09
Social skills variables						
Actor SS	−0.07	0.12	0.24	0.12	0.01	0.10
Partner SS	−0.01	0.12	−0.07	0.12	0.00	0.10
Actor*Partner SS	0.14	0.10	−0.08	0.14	0.02	0.10
Moderation of social skills variables by diagnosis variables						
Actor diagnosis*Actor SS	0.18	0.12	−0.22	0.14	0.05	0.10
Actor diagnosis*Partner SS	−0.04	0.12	0.12	0.14	0.08	0.10
Actor diagnosis*(Actor*Partner SS)	−0.01	0.13	−0.16	0.13	−0.08	0.11
Partner diagnosis*Actor SS	−0.06	0.11	0.07	0.13	−0.10	0.10
Partner diagnosis*Partner SS	0.03	0.12	0.02	0.14	−0.02	0.11
Partner diagnosis*(Actor*Partner SS)	−0.01	0.13	0.21	0.13	0.06	0.11
Moderation of social skills variables by dyad type						
(Actor*Partner diagnosis)* Actor SS	0.08	0.12	−0.14	0.12	−0.04	0.10
(Actor*Partner diagnosis)*Partner SS	−0.00	0.12	0.10	0.12	0.02	0.10
(Actor*Partner diagnosis)* (Actor*Partner SS)	−0.04	0.11	0.11	0.14	0.03	0.10

WRAT-3, Wide Range Achievement Test – 3; SS, Social Skills. All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included.

and these may not have been the best measures to capture meaningful relationships within real-world interaction. Further, using social skill and social cognitive composite scores may have obscured more nuanced effects of specific subcomponent abilities. However, exploratory analyses assessing the effect of performance on individual social cognitive tasks also found few links to interaction outcomes. Further, it is possible that other individual mechanisms not assessed here, such as linguistic abilities and executive functioning, may also have been related to outcomes. Moreover, because some of the effects found in this study were relational and not individual, future studies may seek to move beyond examining individual predictors of social interaction outcomes to instead focus on relational variables, like interpersonal synchrony, compatibility, and affiliation.

Effects may also have been smaller than anticipated because of selection biases in the sample: most autistic participants were students attending college or a professional training program and therefore may have been more independent, intellectually capable, and socially skilled than other autistic adults. Nevertheless, they performed comparably to other autistic samples on measures of social cognition (Bishop-Fitzpatrick et al., 2017; Morrison et al., 2019b), normative social skill (Ratto et al., 2011; Morrison et al., 2017), and social motivation (Sedgewick et al., 2016), suggesting they were largely representative in terms of their measured social abilities. Additionally, because NA participants were mostly psychology students attending a university with a sizeable autistic population, they may have more experience with autism than the general population. Finally, because adequately examining the complicating effects of gender on social interaction outcomes would necessitate a prohibitive increase in sample size and additional dyadic conditions, this study was limited to studying interaction between males. Participants were also disproportionately white because of the racial breakdown of our autism recruitment sources. The lack of gender and ethnic diversity in our sample is perhaps the largest limitation of the

TABLE 15 | Actor-partner interdependence model analyses estimating the combinatorial effects of diagnostic status and social motivation variables on the first impression variables of aggressiveness/dominance, smartness, and liking.

	First-impression outcome variables					
Predictors	**Aggressiveness/Dominance**		**Smart**		**Liking**	
Social motivation predictors	***b***	**SE**	***b***	**SE**	***b***	**SE**
Intercept	1.78	0.12	3.43	0.14	3.34	0.09
Demographic variables						
Actor WRAT	−0.01	0.01	−0.01	0.01	0.01	0.01
Actor race – AfricanAmerican	−0.07	0.19	−0.12	0.24	−0.01	0.14
Actor race – Asian	0.18	0.21	0.22	0.27	−0.26	0.15
Actor race – Other	0.04	0.18	0.19	0.21	0.36*	0.14
Actor age	0.01	0.02	0.04*	0.02	0.02	0.01
Diagnosis variables						
Actor diagnosis	0.00	0.08	0.03	0.08	−0.02	0.07
Partner diagnosis	0.05	0.08	0.00	0.08	−0.02	0.06
Actor*Partner diagnosis	0.08	0.07	0.09	0.09	−0.00	0.05
Social motivation variables						
Actor SM	−0.02	0.01	0.02	0.01	0.01	0.01
Partner SM	0.01	0.01	0.01	0.01	−0.01	0.01
Actor*Partner SM	−0.00	0.00	−0.00	0.00	−0.00*	0.00
Moderation of social motivation variables by diagnosis variables						
Actor diagnosis*Actor SM	−0.00	0.01	0.01	0.01	−0.02	0.01
Actor diagnosis*Partner SM	−0.00	0.01	0.01	0.01	0.01	0.01
Actor diagnosis*(Actor*Partner SM)	0.00	0.00	−0.00	0.00	0.00	0.00
Partner diagnosis*Actor SM	−0.00	0.01	−0.01	0.01	0.01	0.01
Partner diagnosis*Partner SM	−0.00	0.01	−0.01	0.01	−0.01	0.01
Partner diagnosis* (Actor*Partner SM)	−0.00	0.00	0.00	0.00	0.00	0.00
Moderation of social motivation variables by dyad type						
(Actor*Partner diagnosis)*Actor SM	0.00	0.01	0.00	0.01	0.01	0.01
(Actor*Partner diagnosis)*Partner SM	0.00	0.01	0.01	0.01	0.00	0.01
(Actor*Partner diagnosis)*(Actor*Partner SM)	0.00	0.00	−0.00	0.00	0.00	0.00

*WRAT–3, Wide Range Achievement Test – 3; SM, Social Motivation. All continuous variables were grand-mean centered; all categorical variables were effect coded (Diagnosis is coded with NA as the reference group; race is effect coded with white as the reference group). The unstandardized regression coefficients and standard errors come from the corresponding full model in which all of the effects were included. *$p < 0.05$.*

current study. Gender and race are highly salient characteristics within social interactions, and their effects were not explored here. We hope that future studies can leverage more diverse populations to assess how the findings in this study may differ within all female dyads, as well as within cross-gender and cross-racial interactions. In particular, results may be expected to differ for autistic females, who often diverge from autistic males in some aspects of social motivation and behavior (Hull et al., 2017) and tend be evaluated more favorably than autistic males by NA individuals (Cage and Burton, 2019; Cola et al., 2020).

In summary, the current study represents the first comprehensive attempt to directly assess whether and how individual performance on measures of social cognition, social skill, and social motivation among autistic adults predicts their real-world social interaction outcomes with unfamiliar autistic and NA adults. Despite performing lower than NA participants on these measures, autistic adults' performance on each of the three social ability domains was largely unassociated with how autistic adults evaluated—and were evaluated by—their conversation partner. Contrary to prediction, in some cases the social abilities of NA adults were actually more predictive. Taken together, findings from this study raise questions about the predictive utility of standalone measures of social abilities in autistic people for understanding their social interaction difficulties with NA people. Future research should seek to examine and validate measures of real-world social cognition, social skill, and social motivation within an interactive context, and continue to emphasize relational rather than individual predictors of social interaction success.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by The University of Texas at Dallas Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

KM led the design of the study, with contributions from RA and NS. KM gathered and organized the data, with contributions from KD and DJ. KM conducted data analysis in collaboration with RA. KM interpreted the data, with help from KD, DJ, RA, and NS. KM and NS wrote the manuscript, with contributions from RA, KD, and DJ. All authors contributed to the article and approved the submitted version.

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Predicting Uncertain Multi-Dimensional Adulthood Outcomes From Childhood and Adolescent Data in People Referred to Autism Services

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Introduction: Autism spectrum disorder is a highly heterogeneous diagnosis. When a child is referred to autism services or receives a diagnosis of autism spectrum disorder it is not known what their potential adult outcomes could be. We consider the challenge of making predictions of an individual child's long-term multi-facetted adult outcome, focussing on which aspects are predictable and which are not.

Methods: We used data from 123 adults participating in the Autism Early Diagnosis Cohort. Participants were recruited from age 2 and followed up repeatedly through childhood and adolescence to adulthood. We predicted 14 adult outcome measures including cognitive, behavioral and well-being measures. Continuous outcomes were modeled using lasso regression and ordinal outcomes were modeled using proportional odds regression. Optimism corrected predictive performance was calculated using cross-validation or bootstrap. We also illustrated the prediction of an overall composite formed by weighting outcome measures by priorities elicited from parents.

Results: We found good predictive performance from age 9 for verbal and non-verbal IQ, and daily living skills. Predictions for symptom severity, hyperactivity and irritability improved with inclusion of behavioral data collected in adolescence but remained modest. For other outcomes covering well-being, depression, and positive and negative affect we found no ability to predict adult outcomes at any age. Predictions of composites based on parental priorities differed in magnitude and precision depending on which parts of the adult outcome were given more weight.

Conclusion: Verbal and non-verbal IQ, and daily living skills can be predicted well from assessments made in childhood. For other adult outcomes, it is challenging to make meaningful predictions from assessments made in childhood and adolescence using the measures employed in this study. Future work should replicate and validate the present findings in different samples, investigate whether the availability of different measures in childhood and adolescence can improve predictions, and consider systematic differences in priorities.

Keywords: autism spectrum disorder, adult outcomes, early diagnosis cohort, childhood, prediction

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INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder, commonly diagnosed in early childhood and generally thought to have a lifelong impact. It is, however, highly heterogeneous, both among those assessed at any one time and in how individuals develop over time (Howlin et al., 2000; Seltzer et al., 2004; Billstedt et al., 2011; Bishop-Fitzpatrick et al., 2016; Simonoff et al., 2019; Stringer et al., 2020). Given the considerable heterogeneity, it would be clinically helpful for both parents of autistic children and the clinicians they work with to have a clear understanding of what potential adult outcomes could be; both what might be expected with some confidence and where it would be premature to begin to form any expectation. This can be particularly important for individual planning, both in terms of where intervention or support may be required and also to anticipate the potential financial impact of autism, which may be considerable (Buescher et al., 2014). At present, determining prognosis for a young autistic child is a difficult task. Clinicians often rely on translating clinical research which provides meaningful and relevant predictive factors, understanding of average adult outcomes for groups with different characteristics, and important insights gained through the clinician's experience. Additionally, individual opportunities, experiences, and preferences are considered. Limited information is available to guide clinicians and families to determine how the development and phenotypic expression of their child may deviate from other autistic children.

The developmental nature of ASD means that the process of sketching out a long-term prognosis is not a one-time exercise, but one that is refined as a child matures and as the span and depth of measurement increases. This task is made more complicated by the multi-facetted nature of the adult outcome and the increased variety of contexts and resources available to autistic adults, compared to the more structured settings available in childhood. What constitutes a good outcome in adulthood for an autistic child can vary from person to person (Lounds Taylor, 2017; McCauley et al., 2020). Reduction of the severity of symptoms related to ASD can be viewed as both positive or negative (Bagatell, 2010). Poor social functioning has been observed in autistic adults and may lead to reduced quality of life (McCauley et al., 2020) but not always (Billstedt et al., 2011; Howlin et al., 2013). To make prognoses that can capture what a good outcome may look like, we take two approaches. Firstly, we a consider a diverse set of measures to describe outcomes in adulthood. Secondly, we look to create a personalized composite outcome based on the priorities of an individual parent.

This study is one of a series exploring the methods and scope for undertaking individual level outcome prediction for developmental processes. This analysis is based on the Autism Early Diagnosis Cohort (EDX) (Lord et al., 2006, 2020) of children referred for possible autism when aged 2–3 years old. Our previous work using this data has involved grouping participants either using latent class modeling or groups defined by a-priori cut offs IQ and autism diagnosis status (Lord et al., 2020). Latent class modeling is a statistical technique which forms groups of people with similar outcomes, or trajectories of outcomes, across a range of measures. We used a latent class model to reduce the adult outcome, characterized by 15 diverse measures spanning IQ to well-being, to a set of four classes each with a distinctive profile across these measures (Pickles et al., 2020). A second latent class approach to this data involved creating groups based on trajectories of ADHD, anxiety and depression symptoms (McCauley et al., 2020). These different approaches have led to important insights. For example, prediction of latent classes formed from the adult outcome was possible using, in addition to socio-demographic variables, measures of ASD, IQ and a composite of simple functional skills, taken at approximately ages 2, 3, 5, and 9 years of age. The latent classes, however, were heavily influenced by a relatively small subset of easily predicted measures that had been included in the full set of adult-outcome measures. Moreover, parents, clinicians and patients may place greater or lesser importance on different facets of the outcome, and this pattern of relative weight attached to each measure may differ within each of these groups.

This study extends previous work by using a quite different strategy and methods, exploring the impact of extending the span of measures available for prediction into more behavioral and emotional problem domains, and considering prediction from a little closer to the adult outcome by including measures taken beyond childhood and into adolescence. We focus on the prediction of the individual adult outcome measures using regression modeling, which has not been considered in this data before, and then the prediction of composites formed from weighting predictions of individual measures. A notable issue in prediction modeling is the potential for apparent model performance to grossly exaggerate predictive performance in a new sample (Steyerberg and Harrell, 2016). This can be a particular issue when sample sizes are small. To compensate for this we use resampling techniques (bootstrap and cross validation), commonly used in prediction modeling, to provide estimates for model performance taking into account the optimism in apparent measures of model performance (Steyerberg, 2019). A second, related challenge, is that overfitting of the data can lead to poor model performance in new samples. To reduce the risk of overfitting we use LASSO regression, which shrinks parameter estimates toward zero (no association), to prevent associations in the data that exist by chance being modeled (Friedman et al., 2010). Another challenge when predicting outcomes that are measured with some error is that the reliability of the measure can act as a ceiling to predictive performance. Alongside our modeling results we present the limits of model performance that can be expected from reported test-retest reliability of the measure we employ. This allows an assessment to be made as to where we are close to the possible limits of prediction and where it could be possible for improved predictions to be made.

We demonstrate the use of composite outcomes using priorities obtained from parents of autistic children, not involved in the Autism Early Diagnosis Cohort. For this preliminary work, priorities were obtained from parents, rather than the

young autistic people directly. The approach could equally accommodate priorities obtained from the individual themselves. This study differs from earlier work examining predictors of adult outcomes (Magiati et al., 2014; Zimmerman et al., 2018) as we examine the extent to which different measures in the adult outcome can be predicted, rather than which variables are predictors. This work is intended to help frame discussions between clinicians, carers and autistic individuals as to plans and priorities, hopes and evidence-based expectations.

The prediction of individual adult outcome measures may provide insight into measures that could better predict facets of their outcome in adulthood that are currently predicted poorly. It allows us to better identify those aspects of the outcome that are likely determined by early childhood or those predictable only by measures taken in late adolescence. We also identify outcomes that are simply unpredictable, perhaps due to their episodic nature, unreliable measurement, or outcomes which are highly variable due to a large impact of unmeasured, or unobservable, biological or environmental factors.

The potential for developing a prediction tool also raises questions as to the context in which such a tool might be used. This may include the developmental stage of the child, the readiness of the child and parents for information, or the choices they have available or need to make. Incorporating in predictions the autistic individuals' priorities, or their parents priorities, may help reflect this context. Discussion of the clinical implementation of prediction tools requires knowledge of what is and what is not predictable and by when. This study provides a starting point. Work in this area may also be relevant for prioritizing interventions for an individual, or for the development of new interventions, as well as the defining and selection of outcome measures in intervention studies.

MATERIALS AND METHODS

Participants

This analysis uses data from the Autism Early Diagnosis Cohort (Lord et al., 2006). The Autism Early Diagnosis Cohort enrolled 192 participants from North Carolina and Chicago referred for possible autism between age 2 and 3, and 21 referred to the autism program as exhibiting developmental delay. A further 31 participants were recruited from similar sources in Michigan at age 9, with the intention of increasing the sample size for subsequent follow ups. Families and, later, participants (where possible) provided informed consent. The research was approved by institutional review boards from Weill Cornell Medicine,the University of Michigan and UCLA.

The analysis presented in this paper includes the 123 young adults who participated in at least one childhood assessment, and one assessment in adulthood (**Figure 1**). Loss to follow up occurred predominantly due to geographical relocation or losing contact. Twenty four participants (11.3%) declined ongoing participation and were excluded from the analysis. Loss to follow up was associated with race and parental education, with drop out higher for African-Americans and families with the lowest educational levels (Pickles et al., 2020).

Measures and Data Collection

Face to face assessments were undertaken with children and their parents at ages 2, 3, 5 (North Carolina only), 9, 19, 21 (a subset of participants), and 25 years. Further telephone interviews were conducted at age 14, 15, and 17. Assessments were carried out by a team of researchers who had achieved research reliability on the measures administered, led by a Ph.D. level psychologist.

At ages 2–9, and adulthood the severity of autism symptoms was measured using the Calibrated Severity Scores (CSS), calculated from the Autism Diagnostic Observation Schedule (ADOS) (Gotham et al., 2009); Verbal and non-verbal IQ were measured using the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999), Differential Ability Scales (Elliott, 2007) and the Mullen Scales of Early Learning (Mullen, 1995). Daily living skills were measured using the Daily Living Standard score from the Vineland Adaptive Behavior Scales (Sparrow et al., 2005). From age 9, irritability and hyperactivity were measured using subscales of the Aberrant Behavior Checklist (Aman and Singh, 1994); For an overall measure of behavioral problems we used the total problem score taken from the Child Behavior Checklist (CBCL) (Achenbach and Rescorla, 2001) and Adult behavior Checklist (ABCL) (Achenbach and Rescorla, 2003). The Strengths and Difficulties Questionnaire (SDQ) (Goodman, 1997) was completed by teachers about the children in the study at age 14 and 17. In addition to measures used in childhood, adult assessments included the positive and negative subscales from the Positive and Negative Affect Schedule (PANAS-P and PANAS-N) (Watson et al., 1988), the Beck Depression Inventory-II (BDI-II) (Beck et al., 1996); the Well-being Questionnaire (WBQ) (Ryff, 1989); and ordinal assessments of participant's living and work and friendships were made using the Social and Emotional Functioning Interview (SEF-I and SEF-S) (Rutter et al., 1988). Partially completed scales were pro-rated when 80% of items were completed. More details on the measures used and the schedule of assessment are given in **Supplementary Tables 1, 2**.

Eliciting Parent Priorities

To demonstrate how different priorities across outcomes could lead to differences in predictions, we provide predictions weighted with the priorities of two parents of autistic children consulted as part of a parent involvement meeting, arranged as part of a separate study, the priorities for these two parents are labeled parent A and parent B. Questionnaires were completed independently prior to the meeting taking place. Priorities were obtained using a questionnaire which asked parents to allocate points, up to a total of 100, across 10 facets of a child's adult outcome which were then mapped to the outcomes collected in the study (**Supplementary Tables 3, 4**). These priorities are intended to illustrate our approach, not for inferences about more widespread parental priorities.

Sample Size

The available sample size varies across outcomes from 123 for verbal and non-verbal IQ to 91 for the well-being questionnaire. The required sample size for the development of prediction models depends on the number of predictors included in the

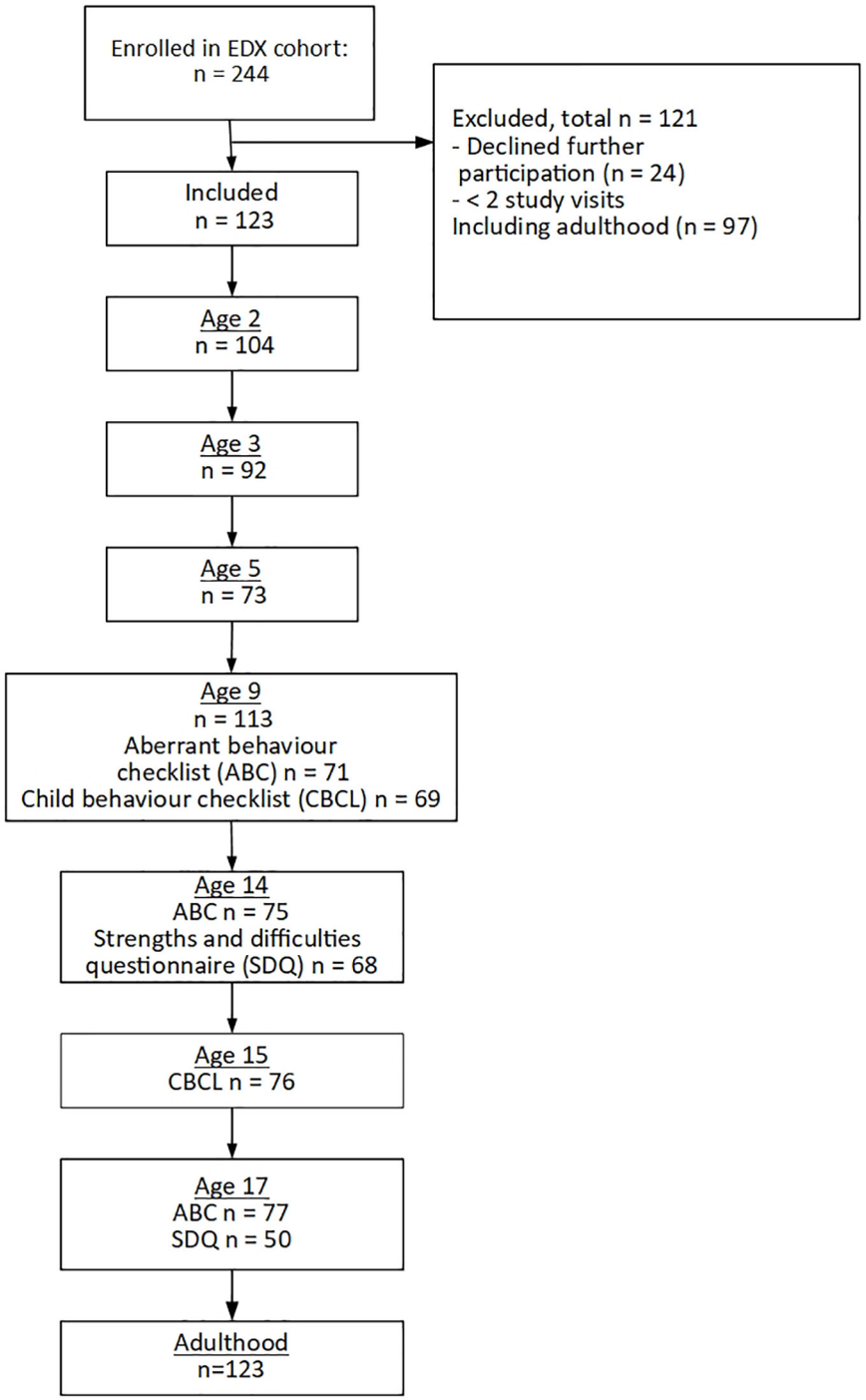

FIGURE 1 | Participant flow through the study.

model and the value of r-squared (Riley et al., 2019). Using Riley et al.'s criteria for linear regression models with 10 predictors (the numbers of predictor we use for models at ages 2–5), and 123 observations the recommended minimum r-squared to reduce the risk of over fitting is 0.44, with 91 outcomes observed outcomes the minimum r-squared is 0.55. For linear regression models fit with 13 predictors (as fitted using post age 9 predictors) minimum r-squared ranges from 0.55 to 0.66 for the available sample size. When the emotion and prosocial subscales of the SDQ are additionally included the minimum r-squared is from 0.6 to 0.7. These r-squared values can be used to guide interpretation of our results, indicating where models may be at

risk of overfitting, which may lead to over-optimistic estimates of predictive performance (r-squared).

Statistical Analysis

For the assessment of predictive performance, statistical analysis was conducted using linear regression for continuous outcomes and proportional odds logistic regression for ordinal outcomes. A separate model was fit for each outcome and at each time point. The analysis for continuous outcomes was repeated using LASSO regression implemented in R using the glmnet package (Friedman et al., 2010), with the tuning parameter selected to minimize the mean squared error using leave-one-out cross-validation. The analysis of ordinal outcomes was not repeated using penalized regression as the low number of events in some categories made it unfeasible to estimate the tuning parameter using cross-validation. Apparent assessments of predictive performance are known to be optimistic as the same data is used to assess predictions as is used to fit the models. To compensate for this, we used leave-one-out cross-validation for continuous outcomes and bootstrap (40 repetitions) for ordinal outcomes (Harrell, 2015). For continuous outcomes predictive performance was measured using R-squared. For ordinal outcomes performance was measured using the generalized c-statistic (Harrell, 2015).

At each time point, the same set of predictors were used for all outcomes. At all ages, models included gender, race, and mothers' education. At ages 2–9, models also included measures from the child's respective age, including verbal and non-verbal IQ, autism symptom severity, daily living skills, and whether the child had a current diagnosis of autism; models beyond age 9 included these variables measures at age 9 as no further assessments of these measures were made before adulthood. Starting at age 9 irritability, hyperactivity, and behavior problems measured using the CBCL were included in the models with new assessments becoming available at ages 14 (age 15 for CBCL) and 17. At each timepoint only the most up to date measurements of predictors were included in the model. Additional models were fit at age 14 and 17 including the pro-social and emotion subscales from the SDQ. **Supplementary Table 5** lists the predictors included at each timepoint.

Prediction Intervals for Weighted Sums of Outcomes

A composite outcome (a single outcome combining all of the adulthood measures) was formed incorporating individual parent priorities. The composite outcomes were calculated by weighting each component by the priority placed on it, then adding together all the weighted components. Prior to summing, outcomes were standardized to have mean zero, and variance one, and where necessary reverse scored so that positive outcomes indicate a more severe impact of autism. Construction of prediction intervals requires consideration of residual standard deviations in addition to standard errors of parameters (Gelman and Hill, 2006). To allow estimation of prediction intervals for weighted combinations of outcomes correlations between residuals for different outcomes needed to be estimated. To estimate these correlations, we used a structural equation model to jointly model all outcomes, with residuals for all outcomes set to be correlated. Standard errors of prediction were calculated for weighted combinations of outcomes, and prediction intervals were calculated assuming outcomes were normally distributed, or for ordinal outcomes, a normal underlying variable of a probit model. Estimation was conducted using weighted least squares, implemented in R using the Lavaan package (Rosseel, 2012).

Missing data in predictors were imputed using k nearest-neighbor imputation, using 5 neighbors (Dahl, 2007). Imputation of missing data was conducted on the whole dataset, prior to splitting for internal validation. In analysis models for single outcomes (those used to assess predictive performance), participants with missing data for an outcome were excluded from the analysis of that outcome (von Hippel, 2007). For joint models, missing outcomes were imputed in the same way as missing data on predictors.

RESULTS

Figure 1 shows the numbers followed up at each time point, and numbers for particular measures when not all participants completed the measure. **Table 1** gives descriptive statistics for predictors measured at enrolment, behavioral measures at age 14, and adulthood outcomes. Summary statistics for all variables, at all timepoints can be found in **Supplementary Table 6**.

Figure 2 and **Table 2** show predictive performance measured using R-squared for continuous outcomes modeled using lasso regression. Results from linear regression were similar and can be found in **Supplementary Table 7**. Test-retest of a measure places a ceiling on our ability to predict it, and meaningful prediction of a wholly unreliable measure is impossible. Unfortunately, we know little about the test-retest performance of many of these measures in samples of this kind, which for many characteristics can vary with age (Rinaldi and Karmiloff-Smith, 2017) and, especially for episodic depression, the performance will vary strongly with the interval between test times. For reference, **Figure 2** therefore displays the upper limits on prediction for different test-retest intra-class correlations (ICC's).

The first panel in **Figure 2** shows results for adult outcomes which were also assessed in early childhood, between ages 2 and 9. For verbal IQ, non-verbal IQ, and daily living skills, predictive performance increases from age 2 to age 9, when the optimism corrected R-squared was 0.88, 0.84, and 0.74, respectively, indicating that precise predictions can be made. The predictive performance at age 9 approaches the test-retest limit of 0.95 for verbal and non-verbal IQ and daily living skills (Balboni et al., 2016; Rinaldi and Karmiloff-Smith, 2017). In contrast to this, predicting autism symptom severity is much more challenging, with only small improvement with age. Our ability to predict adult autism symptom severity improves past age 9 with the addition of adolescent behavioral measures to the model but remains modest.

We found no ability to predict adult behavioral outcomes, such as irritability or hyperactivity from verbal and non-verbal IQ, daily living skills or autism symptom severity measured in

TABLE 1 | Summary statistics for first measure of predictors and outcomes.

Measure	*N*	Median (IQR)/*N* (%)	Range
At recruitment			
Age	123	2.6 (2.2, 2.9)	1.3–11.8
Female—*N* (%)	123	21 (17%)	0–1
Non-Caucasian—*N* (%)	123	21 (17%)	0–1
Maternal education	123	2 (1, 3)	1–5
Autism symptom severity (CSS)	121	7 (3, 9)	1–10
Verbal IQ	123	37 (23, 60)	10–128
Non-verbal IQ	123	75 (54, 85)	13–132
Daily living skills	106	68.5 (61.2, 74)	52–99
Outcomes age 14			
Hyperactivity	75	8 (3.5, 15)	0–31
Irritability	75	5 (1, 13)	0–29
Adult outcomes			
Autism symptom severity (CSS)	118	6 (3, 7)	1–10
Verbal IQ	123	46 (20, 103.5)	2–139
Non-verbal IQ	123	72 (26, 105)	3–133
Daily living skills	123	61 (35.5, 78)	17–112
Hyperactivity	104	4.7 (1.5, 11.6)	0–30.8
Irritability	104	4.7 (1, 9.7)	0–37.5
Behavioral problems (ABCL)	94	53 (48, 57)	25–77
Well-being questionnaire	91	189 (169, 208)	134–248
PANAS-P	92	28 (22.8, 33.5)	12–45.5
PANAS-N	93	15 (12, 21)	10–35.5
Depression (BDI)	92	2.5 (0.5, 7.1)	0–30
Independent living	123	2 (2, 2)	1–3
SEF-I friends	106	2 (0.2, 3)	0–3
SEF-I work	113	4 (2, 6)	1–7

childhood (**Figure 2**, panel 2). From age 9 into adolescence, there is an improvement in predictions of irritability (maximum R-squared 0.40, age 14) and hyperactivity (maximum R-squared 0.43, age 17) due to inclusion of measures in these same domains among the set of predictors that became available as part of the adolescent measurement batteries. Prediction for these outcomes remained modest and fell below what might be the expected test-retest limit. Our success in predicting behavioral problems measured using the ABCL total score (maximum R-squared 0.16) fell below even the modest success that we had with irritability and hyperactivity despite the total score from the related CBCL being included as a predictor in the model. The final panel of **Figure 2** shows that we had minimal success in predicting positive or negative affect (maximum R-squared 0.08 and 0.14, respectively), depression (maximum R-squared 0.01) or well-being (maximum R-squared 0.21) from the data at any age. Inclusion of teacher-reports of behavioral and emotional problems measured using subscales of the SDQ at ages 14 or 17 did not lead to improvement in predictions for any outcome (**Table 2**).

Figure 3 and **Table 3** shows the results for ordinal measures of work, independent living and the SEF-friends scales with improved prediction from age 2 to age 9 as updated measures of verbal and non-verbal IQ, daily living skills and autism symptom severity were added to the model (c-statistics at age 9 of for work, 0.76, independent living and 0.82, friends 0.82 indicating good model performance). There was no improvement in discriminative performance with inclusion of behavioral measures made after age 9.

Parental Priorities

Figure 4 shows the priority profiles for two parents whose priorities we have used to demonstrate our methods for weighted predictions. Priorities greater than 10 indicate a question is given greater importance than if all items were considered equal. Areas of greater importance were depression, behavioral and emotional problems, contentedness and positive emotions. Least priority was given to questions relating to the classic symptoms of autism and having a wide friendship group.

Personalized Composite Outcomes

Personalized composite outcomes were created by summing individual outcomes each with a weight. The weights were derived from the parent priorities questionnaire results. **Figure 5** showed predictions, and 95% prediction intervals for a hypothetical child, aged 15, with scores across all predictors at the 25th centile for impact of autism of the imputed data used to fit the model (Age 9, Verbal IQ = 101, Non-verbal IQ = 95, daily living skills = 45, autism symptom severity (CSS) = 4, Age 14 irritability = 1, hyperactivity = 4, Age 15, CBCL = 50).

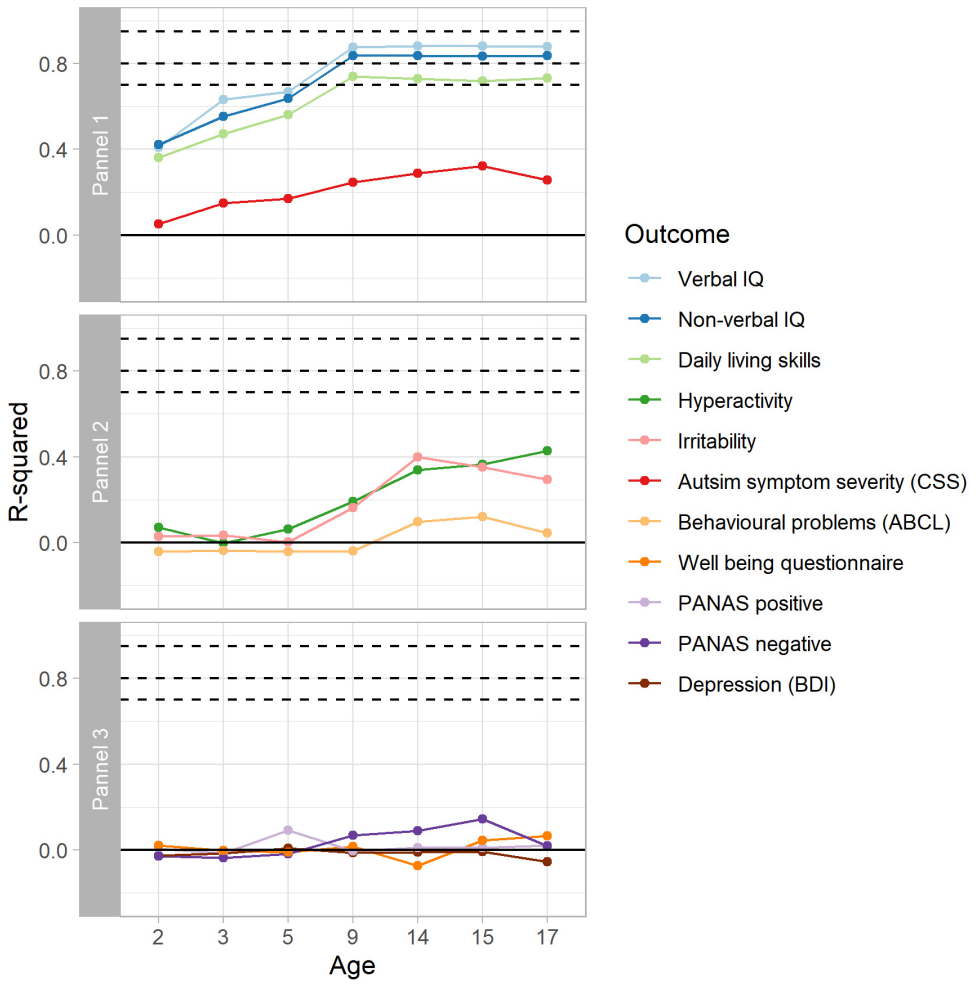

FIGURE 2 | Optimism corrected predictive performance for continuous outcomes modeled using Lasso regression. Dashed lines show limits of predictive performance for test-retest ICCs of 0.95, 0.8, and 0.7. For verbal IQ, non-verbal IQ, and daily living, the results reach close to the test-retest limits for the outcome measures.

TABLE 2 | Optimism corrected predictive performance for continuous outcomes, modeled using lasso regression.

Outcome	2	3	5	9	14	14*	15	17	17*
Verbal IQ	0.41	0.63	0.67	0.88	0.88	0.88	0.88	0.88	0.88
Non-verbal IQ	0.42	0.55	0.64	0.84	0.84	0.85	0.83	0.84	0.83
Daily living skills	0.36	0.47	0.56	0.74	0.73	0.74	0.72	0.73	0.75
Hyperactivity	0.07	0	0.06	0.19	0.34	0.37	0.37	0.43	0.43
Irritability	0.03	0.03	0	0.16	0.4	0.35	0.35	0.29	0.28
Autism symptom severity	0.05	0.15	0.17	0.25	0.29	0.23	0.32	0.26	0.25
Behavioral problems (ABCL)	−0.04	−0.04	−0.04	−0.04	0.1	0.07	0.12	0.04	0.05
Well-being questionnaire	0.02	0	−0.01	0.01	−0.07	0.1	0.04	0.06	0.21
PANAS positive	0.01	−0.02	0.09	0	0.01	0.08	0.01	0.02	−0.04
PANAS negative	−0.03	−0.04	−0.02	0.07	0.09	0.05	0.14	0.02	0.06
Depression (BDI)	−0.03	−0.02	0.01	−0.01	−0.01	−0.02	−0.01	−0.06	−0.04

**At age 14 and 17 additional analysis were conducted including the pro-social and emotion subscales from the strength's and difficulties questionnaire.*

The predictions of individual outcomes (**Figure 5**) showed that across the IQ measures and daily living skills it was most likely that the child would be less impacted by symptoms of autism in adulthood. Predictions for irritability, hyperactivity, and autism symptom severity showed that while it was likely that the impact would be less severe, there was still the possibility of a

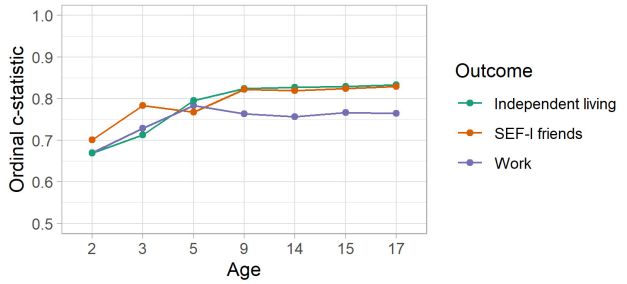

FIGURE 3 | Optimism corrected predictive performance for ordinal outcomes.

more profound effect of autism symptoms. For other outcomes, predictions close to zero and wide prediction intervals showed that predictions added little beyond describing the distribution in the population. These are in line with what we expected from our assessments of predictive performance.

Figure 6 showed predictions for a parent-specific composite outcome formed by weighting the same set of predictions of individual outcomes by the priority sets of different parents. Differing parent priorities led to different predictions of their composite with varying levels of confidence in prediction. For Parent A who placed relatively more weight on depression and well-being, areas that we were unable to predict well, the prediction interval (-1.30 to 0.4) was wide, and included some possibility of a greater than average impact of autism symptoms. For Parent B the 95% prediction interval (-1.26 to 0.06) largely excluded the possibility of a more severe than average impact of symptoms because Parent B placed greatest priority on items related to behavioral and emotional problems and practical tasks, for which by age 15 we achieved better prediction performance.

DISCUSSION

Discussions that patients, parents and clinicians have about priorities and planning in autism vary greatly. The structured evidence that we have for what is and what is not predictable is poor, leaving clinicians to make judgements based on their personal experience. Personal communication from some parents suggest that there may be a small minority of clinicians who feel very confident in what should be considered important, their ability to predict these, and who delivers their predictions with apparent certainty. Perhaps the principal message of this paper is that there is not necessarily the evidence to support such a practice as there can be differences in what parents consider important, and for some aspects of the adult outcome, prediction can be extremely challenging.

Based on assessments of verbal and non-verbal IQ, daily living skills and autism symptom severity it is possible to make good predictions at age 9 of a child's adult IQ and adaptive functioning. Predictions of friends, work and living situation are also possible. The importance of verbal and non-verbal IQ and symptom severity in making long term prognosis for autistic children is consistent with results from previous systematic reviews (Magiati et al., 2014; Zimmerman et al., 2018). However, these measures are insufficient to predict adult assessments of irritability and hyperactivity, and it is only with the inclusion of adolescent measures of these outcomes that predictions of these outcomes are possible with modest certainty. No measurements of irritability or hyperactivity were made prior to age 9, however, the relatively poor predictions possible with assessment of these measures at age 9 indicate earlier assessments would be unlikely to contribute to improved predictions. Predictions of the severity of autism symptoms measured using the CSS improved as childhood measures of IQ, daily living skills and autism symptom severity were updated, and with the addition of behavioral measures made in adolescence. Predicting other measures which make up the adult outcome proved extremely challenging. Based on the available data it is not possible to predict behavioral problems measured using the ABCL, adult well-being, depression, or positive or negative emotions with any certainty. While it may be considered that accurate prediction is the critical goal for planning, providing an evidence-based indication of the uncertainty of predictions is equally important. This avoids unsubstantiated over-generalized and deterministic views of the future, maintaining scope for appropriate hopes and ambitions while avoiding both the dispiriting effects of failing to achieve near-certain unrealistically positive goals and failing to grasp within-range opportunities through unjustified pessimism.

Predictions of the composite of all outcomes formed using parental priorities are more precise and have prediction intervals that encompass a range of outcomes closer to the average than predictions of the individual components. This is due to the weak correlation among the prediction errors across the outcome profile. This leads to predictions closer to the average because a more- extreme- than- expected- outcome in one aspect of the outcome profile does not mean that a more- extreme- outcome should be expected in another aspect of the profile. Therefore, when combining predictions for many outcome measures, a less extreme outcome will be predicted.

The priorities elicited from parents show that it is possible to incorporate these views into predictions of future outcomes.

TABLE 3 | Optimism corrected performance for ordinal outcomes modeled using the proportional odds model.

Outcome	2	3	5	9	14	14*	15	17	17*
Independent living	0.67	0.71	0.79	0.82	0.83	0.81	0.83	0.83	0.82
SEF-I friends	0.7	0.78	0.77	0.82	0.82	0.83	0.82	0.83	0.82
Work	0.67	0.73	0.78	0.76	0.76	0.76	0.77	0.76	0.75

**At age 14 and 17 additional analysis were conducted including the 5 subscales from the strength's and difficulties questionnaire.*

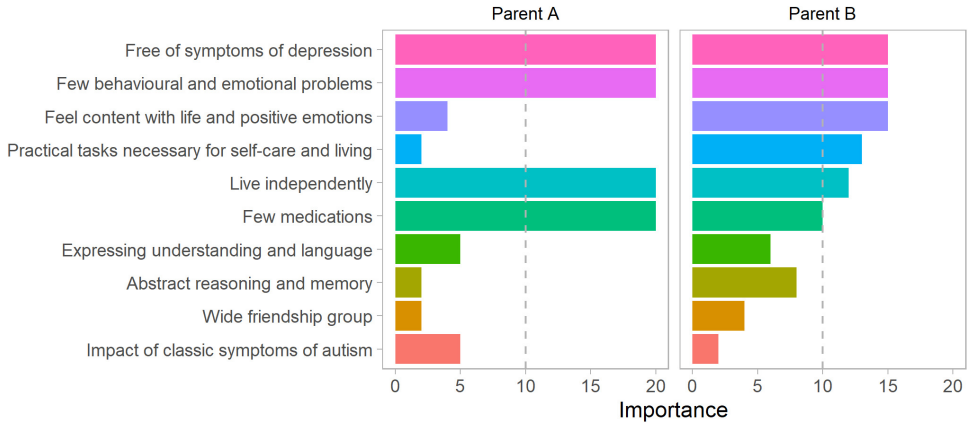

FIGURE 4 | Individual parent priority profiles for two parents. This figure shows outcome priorities elicited from two parents of autistic children, labeled parent **(A)** and **(B)**. The dashed vertical line shows the priority that would be given if all outcomes were considered equal.

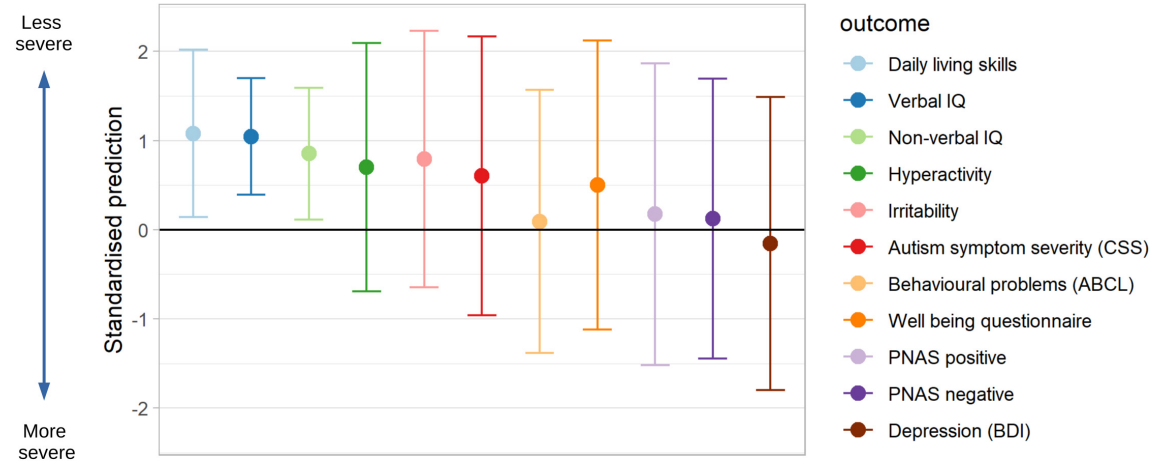

FIGURE 5 | Predictions of continuous outcomes for a child, age 9, at 25th centile for impact of autism. Point predictions and 95% prediction intervals for a child with scores at the 25th centile for impact of ASD on predictors (Age 9 Verbal IQ = 101, Non-verbal IQ = 95, daily living skills = 45, CSS = 4, Age 14 irritability = 1, hyperactivity = 4, Age 15 CBCL = 50). Higher scores indicate a less severe impact of ASD.

Although not generalisable to the wider population, the parent priorities elicited in this study show that there is diversity in what parents of autistic children consider important in relation to adult outcomes. This is consistent with observations that what makes up a good outcome may be personal (Lounds Taylor, 2017). The aspects of the outcome related to symptoms of depression, contentedness with life and positive emotion, where we had the greatest challenges in making predictions, were amongst the areas given highest priority on average by parents. There is no need to assume that the that priorities need be fixed for a particular child. It would be very reasonable to expect priorities to reflect current concerns more sharply than those associated with a hard to imagine domain of a distant future life. While recalculation and discussion with updated weights may be appropriate, consideration might be given to a series of such discussions over the years, but with each being concerned with outcomes more proximal in time and developmentally closer to the contemporaneous lived experience.

The models developed in this study are highly informative with regard to how different facets of the adult outcome can be predicted and how predictions change over time, but further external validation, in new data, is required before the results should be used to make personalized predictions in clinical practice (Steyerberg and Harrell, 2016). The modest sample size available in this study also means that there was the potential for overfitting. This could have led to optimistic estimates of model performance for irritability, hyperactivity and autism symptom severity (measured using the CSS). The risk of overfitting from more complex models also lead to us adopting a conservative analysis strategy, using a relatively simple modeling approach. A larger sample size may support more complex modeling which could improve prediction accuracy. The findings are based on a single study, carried out in a particular context;

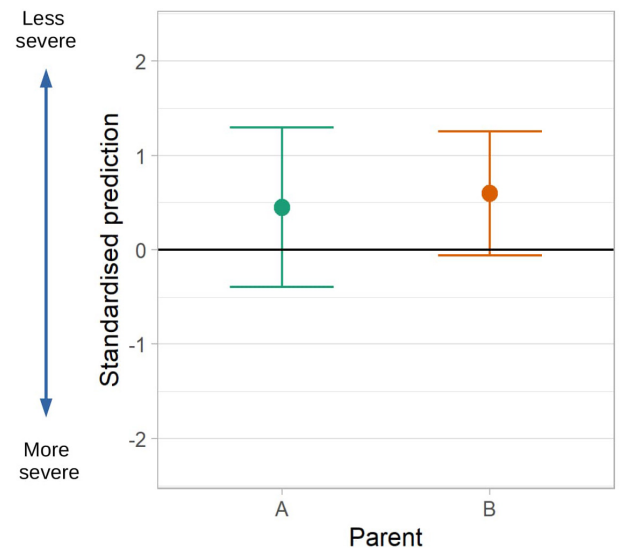

FIGURE 6 | Predictions of a composite outcome formed with different parent priority profiles. Point predictions and 95% intervals for a child with scores at the 25th centile for impact of ASD on predictors (Age 9 Verbal IQ = 101, Non-verbal IQ = 95, daily living skills = 45, CSS = 4, Age 14 irritability = 1, hyperactivity = 4, Age 15 CBCL = 50). Higher scores indicate a less severe impact of ASD. Predictions of the composite outcome are similar for both parents, however based on Parent **(A)** priorities there is more certainty in the predictions than for Parent **(B)**.

predictive performance in areas we found it to be poor may improve if additional measures were included in childhood or adolescence. A change in the availability of effective therapies could also lead to different results. The modeling approach we took means our results should be interpreted in the context of a child undergoing two assessments—one between the age of 2 and 9, and a second in adolescence. A different approach, incorporating repeated measures of the same predictor in models would be required to model the effect of a more intense program of assessment.

Future work should consider replicating these findings in other datasets, investigating whether different sets of measurements can improve predictions in areas we found challenging, and considering analysis approaches which incorporate repeated measures of predictors. Further development of prediction models would also benefit from a participatory approach where autistic people and parents of autistic children are involved in all stages of the research. Predictions might also change as the life-opportunities of autistic adults change, given substantial geographical and temporal variability. Additional work is also clearly required to better understand what parents of autistic children, and the children themselves, want and want to know. In addition, differences in priorities between autistic individuals, their families, and the professionals they work with, and for children of differing capabilities, developmental stage and in different settings of cultural expectations and opportunity could be examined.

CONCLUSION

Assessments in childhood can lead to good predictions of cognitive ability, daily living skills, and social functioning. Predictions improve with age up to age 9. Prediction of the severity of autism symptoms in adulthood improved throughout childhood and adolescence, but predictions remained weaker than for cognitive ability or adaptive functioning. For behavioral aspects of the adult outcome, prediction is only possible with assessments made in adolescence and even then remain uncertain. For aspects of the adult outcome relating to mental health and well-being, prediction was extremely difficult at any age. One feasible way to summarize multi-faceted adult outcomes is to combine different adult outcomes measures into a single composite based on the individual or consensus priorities of parents, clinician and autistic individuals. We are continuing to work on the development of methodology and tools that can facilitate the process of discussing the future and its implications for current priorities and planning.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because they contain personal sensitive data concerning the health of participants. Requests to access the datasets should be directed to GF, gordon.forbes@kcl.ac.uk.

ETHICS STATEMENT

The Early Diagnosis Cohort was reviewed and approved by the Weill Cornell Medicine IRB and IRBs at the University of Michigan. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

GF conducted the statistical analysis, wrote the first draft of the manuscript. AP conceived the idea for the analysis provided input into methods used. CL has led the study since its inception and RE lead the data collection in adulthood. All authors contributed to further drafts of the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.594462/full#supplementary-material

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ORIGINAL RESEARCH
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Visual Noise Effect on Contour Integration and Gaze Allocation in Autism Spectrum Disorder

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Contradictory results have been obtained in the studies that compare contour integration abilities in Autism Spectrum Disorders (ASDs) and typically developing individuals. The present study aimed to explore the limiting factors of contour integration ability in ASD and verify the role of the external visual noise by a combination of psychophysical and eye-tracking approaches. To this aim, 24 children and adolescents with ASD and 32 age-matched participants with typical development had to detect the presence of contour embedded among similar Gabor elements in a Yes/No procedure. The results obtained showed that the responses in the group with ASD were not only less accurate but also were significantly slower compared to the control group at all noise levels. The detection performance depended on the group differences in addition to the effect of the intellectual functioning of the participants from both groups. The comparison of the agreement and accuracy of the responses in the double-pass experiment showed that the results of the participants with ASD are more affected by the increase of the external noise. It turned out that the internal noise depends on the level of the added external noise: the difference between the two groups was non-significant at the low external noise and significant at the high external noise. In accordance with the psychophysical results, the eye-tracking data indicated a larger gaze allocation area in the group with autism. These findings may imply higher positional uncertainty in ASD due to the inability to maintain the information of the contour location from previous presentations and interference from noise elements in the contour vicinity. Psychophysical and eye-tracking data suggest lower efficiency in using stimulus information in the ASD group that could be caused by fixation instability and noisy and unstable perceptual template that affects noise filtering.

Keywords: contour integration, visual perception, ASD, neural noise, external noise, eye movements

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INTRODUCTION

Atypical processing of low-level sensory information has been reported in Autism Spectrum Disorder (ASD) (Dakin and Frith, 2005; Simmons et al., 2009) in addition to impaired social and higher-level cognitive abilities, restricted and repetitive behaviors. The significance of sensory symptoms, like abnormal reactivity to sensory stimuli manifested as either hyper- or

hypo-sensitivity is emphasized by their inclusion in the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association [APA], 2013).

One of the most notable examples of altered perception in ASD is the compromised processing of social stimuli such as faces. Along with the assumption that the impairment derives from a pervasive problem in social interaction and motivation, several studies are suggesting that the visual perceptual alterations may contribute to the difficulty with face processing as well (reviewed by Behrmann et al., 2006). A possible mechanism of the suboptimal face processing could be connected to the well-described diminished ability in ASD to group local visual elements that are presented in different parts of an image into a global percept despite the enhanced processing of visual details (Behrmann et al., 2006; Happé and Frith, 2006).

Different neurophysiological studies explored the question of how local signals are integrated across space to generate global percepts. The data obtained are interpreted as evidence that horizontal, feedforward, and feedback connections between neurons in the visual system, particularly in the primary visual cortex (V1), are responsible for the visual integration (e.g., Kapadia et al., 2000; Angelucci et al., 2002; Nurminen and Angelucci, 2014).

At the psychophysical level, the ability to group or integrate local visual elements has been often explored by contour integration studies that involve the detection of a contour consisting of Gabor elements embedded among a background of randomly oriented Gabors (e.g., Field et al., 1993; Jachim et al., 2015). The target contours could represent a single line, named as an open contour (Field et al., 1993), or a predetermined shape, closed contour (Jachim et al., 2015). Kovács and Julesz (1993) first reported that it is much easier to detect closed than open contours and their finding was repeated in later studies (Mathes and Fahle, 2007; Gerhardstein et al., 2012; Jachim et al., 2015). In order to explain this facilitated detection, it was suggested that in an early vision a synergetic process exists (Kovács and Julesz, 1993) or a separate mechanism that is sensitive to the detection of closed contours (Gerhardstein et al., 2012). Mathes and Fahle (2007) suggested that closed contour facilitation may occur at both early visual areas which are responsible for local orientation information processing and at higher visual areas (the lateral occipital complex) which process the global shape of the contour.

An important factor that determines the contour integration ability is the alignment of the elements along "the path" (Field et al., 1993). The detection of contours is diminished when the relative orientation or jitter of adjacent Gabor elements is increased (Field et al., 1993; Jachim et al., 2015).

The results, obtained in the studies that compare contour integration abilities in ASD and typical development (TD), are contradictory. Del Viva et al. (2006) found similar spatial integration performance between children with ASD and TD when detecting a circle embedded in noise. The elements of both the circle and the noise were Gabor patches presented for 1 s on a computerized display. The authors interpreted these findings as a demonstration of preserved early perceptual integration. Kemner et al. (2007) applied a card-based version of the contour integration task with closed contour stimuli and over a second-long presentation time. They similarly found normal performance in the group with a pervasive developmental disorder compared to the control group.

Contrary to these results, contour integration ability was challenged in later studies. By applying an electrophysiological paradigm Pei et al. (2009) searched for neural correlates of the local visual signals integration in a group of low functioning children with ASD and an age-matched control group. The stimuli were Gabor elements that alternated every 500 ms forming circular contours or random patterns. It was found that the 3rd harmonic response was absent in the group with autism in contrast to the control group. The authors interpreted this finding as a neural correlate of highly specific deficiencies that could be connected to some deficits in ASD like face avoidance or reading abnormalities. Evers et al. (2014) compared the identification of gradually appearing contours by aligning local Gabor elements toward randomly oriented Gabor elements. The result showed that the identification performance of the children with ASD was slower and less accurate than that of the controls with TD, especially when more complex contours were shown. The results were interpreted as evidence of an impaired relationship between local-global and bottom-up-top-down processes in autism. Hadad et al. (2019) also reported slower and less accurate responses in the ASD group than in the TD group in identifying contours based on everyday objects. However, the authors suggested that the group differences could indicate known differences between the groups in response times and general tolerance to noise, rather than in the mechanism of spatial integration.

It seems that at least several factors could be responsible for the contradictory results between the different studies on contour integration ability in autism. Jachim et al. (2015) suggested that the peculiarities of the atypical contour integration in ASD became obvious mostly in cases of object identification instead of detection of simple shapes. In their study, Jachim et al. (2015) found less improvement in contour detection between open and closed contours in adults with ASD compared to a control group with TD, although there was not a group difference with either open or closed stimuli. In contrast to the last finding in the newest study on this topic (Gowen et al., 2020) better perceptual performance for ASD compared to the TD group was observed for the open stimulus in addition to the replication of the reduced closure effect. As possible explanations, the authors discussed several possibilities: the involvement of autistic participants with an enhanced perceptual ability, the difference in the number of Gabor elements in the open contour, as well as the possibility for more eye movements toward the contour made from the autistic group and thus improving the contour detection because the central instead of the peripheral location improves contour integration (e.g., Hess and Dakin, 1997; Nugent et al., 2003). However, Gowen et al. (2020) suggested that eye movements could hardly influence their results since the deviations from the fixation point greater than 2.5° from the center were removed and there was no improvement in performance between short and long stimulus duration.

Long stimulus duration is indicated as a possible factor that could hide any differences in contour integration since people

with ASD may need more time to discern the figure (Jachim et al., 2015). Based on a meta-analysis Van der Hallen et al. (2015) concluded that global-order perception is slower in ASD than in TD. However, the results of Gowen et al. (2020) showed a similar reduced closure effect in the ASD group compared to TD at short (150 ms) and long (500 ms) stimulus presentation times, thus rejecting the possible role of slower global processing. Nevertheless, there are still considerations that participants with ASD could apply a different strategy in contour integration tasks (reviewed by Jachim et al., 2015) or in face recognition tasks (e.g., Deruelle et al., 2004; Ashwin et al., 2006), especially at longer stimulus durations.

Generally speaking, it has been assumed that two types of determinants govern human signal-detection responses: external and internal (e.g., Burgess and Colborne, 1988). While external determinants are connected to the nature of the signal, the noise, and the task, variability in the internal determinants is commonly attributed to internal noise. Internal noise influences the nervous system at each level starting from the perception of sensory signals to the generation of motor responses (Faisal et al., 2008) and has been reported in sensory and motor systems of individuals with autism (Simmons et al., 2009; Dinstein et al., 2015). Higher neural variability in visual, somatosensory, and auditory modality was demonstrated in functional magnetic resonance imaging (fMRI) studies by poor evoked response reliability when comparing cortical response amplitude and consistency across trials (Dinstein et al., 2012) or by greater intra-individual variability in the sensory-evoked fMRI responses (Haigh et al., 2015). In support of the assumption about the increased neural noise in ASD are also results from electrophysiological studies. Milne (2011) observed significantly greater intra-participant electroencephalogram (EEG) variability and lower inter-trial α-band phase coherence in ASD individuals than in neuro-typical matched controls. Weinger et al. (2014) reported lower signal-to-noise ratios and deficits in low-contrast responses at the stimulus frequency of 12.5 Hz in the ASD group compared to the TD group. Increased inter-trial variability in ASD that resulted in reduced P100 amplitude was recently described by Kovarski et al. (2019).

Psychophysical features in ASD such as high visual motion coherence thresholds (Milne et al., 2002) and broad tuning of auditory filters (Plaisted et al., 2003) could be explained by high levels of noise in neural networks as suggested by Baron-Cohen and Belmonte (2005). The signal-to-noise ratio could be reduced if a network is overconnected and sensory inputs evoke atypically large activations for both attended and unattended stimuli resulting in an overall unselective increase of activation (Belmonte et al., 2004). However, the results of other studies (Brock et al., 2002; Just et al., 2004) imply diminished connectivity. Baron-Cohen and Belmonte (2005) suggested that this contradiction could be explained by the possibility that the high connectivity within local networks could develop together with atypically low computational connectivity with other regions.

Excessively high levels of neuronal noise could be generated at both the neural network level and at the single-cell level. Increased inner noise may result from high variability of neuronal activity in peripheral receptors (Schneeweis and Schnapf, 1999, 2000; Faisal et al., 2008), or synaptic transmission variability due to the probabilistic nature of the neurotransmitter release and the variable timing and amplitude of the post-synaptic response (Ribrault et al., 2011). Mechanisms that target excitatory and inhibitory synapses, and mechanisms that target intrinsic neuronal excitability support the balance between excitation and inhibition that could be probably compromised in autism (Turrigiano, 2011). Persico and Bourgeron (2006) reviewed genetic, epigenetic, and environmental factors that could contribute to autism. The authors suggested several major pathways that are concerned in ASD pathogenesis: altered cell migration, the glutamate–GABA equilibrium, synapse formation and maintenance, as well as dendritic morphology. Single-neuron sensory responses depend on the states of their neural networks and changes in levels of attention and excitement (Fontanini and Katz, 2008). At the neural network level, variability can be increased due to disturbances of excitation/inhibition balance through increased levels of excitatory inputs (Rubenstein and Merzenich, 2003; Trakoshis et al., 2020) as well as by continuous interaction and competition between functional brain networks (Kelly et al., 2008). Network inefficiencies could be connected to deficits in connectivity related to low-level processing and could potentially affect higher-level cognitive processes and social behavior (Lewis et al., 2017).

However, it should be noted that the question of the higher internal noise in ASD is still disputable. Butler et al. (2017) observed similar levels of variability in visual and somatosensory evoked EEG using high-density mapping in individuals with ASD and TD. The comparison of the magnetoencephalographic response to passive tactile stimulation failed to show higher variability in the ASD group than in the group with TD (Coskun et al., 2009). A psychophysical study on motion integration applying the equivalent noise approach, which uses different quantities of external noise added to the stimulus, (Manning et al., 2017) revealed enlarged sampling in children with ASD for motion information but no convincing evidence for abnormal levels of internal noise. Davis and Plaisted-Grant (2015) suggested that symptoms of ASD could be explained by reduced instead of increased endogenous noise, which is probably a function of abnormal brainstem activation. Low internal noise would lead to increased detection and discrimination in ASD. However, a low-noise brain will not gain benefits of noise in neural networks and may fail to generalize learning from one context or stimulus to others; become "stuck" in a certain mode of thought or action; may not respond reliably to a stimulus across presentations.

Concerning the external determinants of the signal-detection response, it should be noted that in most contour integration studies, external noise is inherent to the stimuli since the target contour is constructed from elements positioned among many similar "noise" elements. The physical randomness in the external environment could induce perceptual variability (Bialek, 1987). Moreover, Osborne et al. (2005) supposed that even the variability in movements could result from errors in the sensory estimates of the external parameters defining the appropriate action rather than by noise in the motor system itself. The irrelevant noise

in the sensory signal is usually excluded through a process of external noise filtering by an appropriate perceptual template, thus diminishing the negative effects of added noise (Lu and Dosher, 2008; Park et al., 2017). The ability to filter the noisy signals would maintain our perception stable, while suboptimal external noise filtering would reduce perceptual efficiency.

The ability to filter the noisy signals is diminished in ASD (Park et al., 2017). Manning et al. (2015) suggested that segregation of signal from noise could be a limiting factor for individuals with autism across a range of motion processing tasks. Children with autism showed enhanced motion integration compared to typical children, but similar performance in the motion coherence task, which requires reporting the direction of coherently moving dots among randomly moving noise dots. These results were interpreted as an implication that the motion coherence thresholds in autism may be affected by diminished discrimination of signal from noise. The authors suggested that seemingly advantageous increased integration may lead to feelings of "sensory overload" in children with ASD. Sanchez-Marin and Padilla-Medina (2008) found that children with autism detected a simple visual signal, still or in motion, embedded in Gaussian noise, significantly worse than children with TD. The authors concluded that this result is not connected to a limited ability to detect simple visual stimuli in autism because the stimuli used in their study were not easy to detect, even for TD children. Most probably, the overwhelmed or disturbed children's ability to process the visual information due to the background noise and motion was responsible for the results. Except for the additive noise, the induced internal noise (Burgess and Colborne, 1988) proportional to the external-noise spectral density could also limit behavioral performance. It is possible that the induced internal noise increases more strongly for the observers with ASD than for observers with TD, and this could lead to anomalous processing of the detected information (Sanchez-Marin and Padilla-Medina, 2008). Zaidel et al. (2015) found that the addition of stimulus noise to visual motion through a cloud of dots affected significantly more the perception of adolescents with ASD than controls despite that the results of both groups were similar without noise. The authors interpreted these results as increased sensitivity to sensory noise and less use of prior knowledge in ASD.

The perceptual efficiency could be reduced by both poorer external noise filtering and excessive neural variability levels referred to as neural or inner noise (Park et al., 2017). Results of Park et al. (2017) demonstrated that both factors are affected in ASD: the internal noise is elevated, and the external noise filtering is diminished. A complicating factor is the difficulty to separate the effects of diminished external noise filtering and increased internal neural noise. External sensory stimuli being naturally noisy could influence the internal noise and could increase trial-to-trial variability at the first stage of perception during the processes of conversion into a chemical or mechanical signal as well as during the following processes of amplification and transduction of the sensory signal and conversion it into an electrical impulse (Faisal et al., 2008).

The aim of the present study was to explore the limiting factors in contour integration processing in ASD. We tried to evaluate the potential role of elevated internal noise and a noisy or variable perceptual template for contour detection using psychophysical methods and eye-movements recording. To achieve this goal, we suggested a stimulation that differs in several aspects from the typical studies on contour integration. A significant difference is that while in the other studies, the background elements are distributed pseudo-randomly on a square grid, in our study, all elements are positioned precisely at the intersection points of a regular hexagonal grid. Therefore, their centroids are aligned with the grid, and no positional information distinguishes the contour elements from the background noise. The observers had to detect a tilted straight contour aligned with a virtual line from the grid among randomly oriented similar elements. The position of the contour (when present) was fixed. We varied the contour strength by changing the orientation of the contour elements by variable amount keeping the mean contour orientation the same but altering the orientational variability. The increased orientation variance represents the external noise added to the contour. This manipulation effectively changes the similarity between the contour and background elements. We limited the stimulus presentation to 200 ms to minimize the possible impact of uncontrolled eye movements and to restrict the possibility of searching behavior. However, we registered the observers' eye positions during stimulus presentation to obtain information on whether their gaze positions vary with the stimulus characteristics.

We presented the stimuli with the same orientational variability in blocks. This would allow the observers to obtain a proper template for each contour strength. While the observers could not change their gaze allocation during the short stimulus presentation, they could have moved their eyes during the fore-period due to either fixation instability or differences in the template. The fixation instability should be independent of the stimulus while the stimulus-dependent gaze shifts and their variability can provide a measure of template stability. In addition, we used the double-pass paradigm (Burgess and Colborne, 1988) at two noise levels – low and high. This paradigm is regarded as the most appropriate for evaluating the factors limiting human performance. The methodology allows partitioning the behavioral variability in correlated and uncorrelated factors. The correlated factors are related to the stimulus variability, while the uncorrelated ones are due to the internal noise that randomly changes. The double-pass paradigm consists of repeating the stimulus sequence and comparing the agreement between the responses to the same stimuli in the two repeats and the accuracy of performance. If no internal noise limits the performance the responses in the two repeats should be the same, whereas the accuracy will be determined by the stimulus variability. At low levels of stimulus variability (low levels of external noise), the performance will be limited by the additive internal noise. At high levels of external noise, the contribution of the additive internal noise becomes negligible and the behavioral performance is limited by stimulus-dependent (multiplicative) noise or by suboptimal computations like missing important stimulus features or using irrelevant stimulus characteristics i.e., the irrelevant information is not filtered. The double-pass paradigm allows the evaluation of the

ratio of the internal to external noise. Therefore, it permits comparisons of the internal noise levels between the ASD and TD groups at the same external noise level.

We tried to restrict the confounding effect of some of the factors mentioned above. To avoid an influence from hierarchically higher areas like the lateral occipital complex (Murray et al., 2002; Gilad et al., 2013), we decided to use open contours instead of closed contour stimuli. We tried to make the participants' task as simple as possible to prevent the task difficulty effect on the results. To prevent the participants with ASD from using a different strategy to determine contour presence or absence, the stimulus duration in our experiments was limited to a short presentation time. To cover a representative part of the autistic spectrum, we tried to include in our study children and adolescents with a wide range of IQ and different proximity to the ASD cut-off as calculated by ADI-R.

We expect that if the participants with ASD have higher levels of additive internal noise or could not filter the background noise, their performance would be worse than that of the participants with TD, even when no external noise is added to the contour. If participants with ASD have higher stimulus-dependent or induced noise, they will show reduced agreement between the responses in the two repeats at the higher level of external noise in the double-pass experiment. If the response time in ASD varies in a stimulus-dependent manner, this will imply that the potential differences between the ASD group and the TD group are not connected only to the preparation and the execution of the motor response. Stimulus-dependent changes in the response time may reflect the different time needed for stimulus encoding at the different levels of external noise or the difference in the rate of evidence accumulation for a particular response choice due to task difficulty changes. If the gaze positions vary with the added external noise, this might be regarded as a noisy or variable template for contour detection at different noise levels.

MATERIALS AND METHODS

Participants

Sixty children and adolescents participated in the study: 28 in the ASD group (4 were later excluded from the analysis) and 32 in the TD group. The participants were recruited via the Sofia Center for Social Rehabilitation and Integration–autism spectrum priority, the Regional Center for Support of the Inclusive Education Process-Sofia-city, Regional Department of Education–Sofia city and through community organizations, parental associations, and professionals (psychologists, speech therapists, child psychiatrists, etc.).

Brief interviews and a developmental questionnaire (filled by parents) ensured that none of the participants in the study have a history of previous neurological or psychiatric disorder (other than ASD in the experimental group), head trauma, current psychoactive medication, and the presence of a visual impairment that could interfere with the performance of tasks. No learning or language difficulties were reported for the TD group. Wechsler Intelligence Scale for Children–Fourth Bulgarian Edition (WISC–IV BG, 2015; Wechsler, 2003) was administrated for both groups, resulting in Verbal Comprehension Index (VCI), Perceptual Reasoning Index (PRI), Working Memory Index (WMI), Processing Speed Index (PSI), and Full-Scale IQ (FSIQ) (see **Table 1**).

At first, the ASD group consisted of 28 children and adolescents, 4 of whom were unable to perform the experimental task adequately and their data were excluded from the analysis. Thus, the final sample included 24 participants with ASD (16 boys, and 8 girls; mean ± SD [range] age = 11.6 ± 2.4 [8–16] years). All of them had already been diagnosed with a pervasive developmental disorder (including Autism, Asperger's syndrome, and ASD) according to ICD-10 (International Statistical Classification of Diseases and Related Health Problems 10th Revision, 1990) criteria. For the study, the diagnosis was confirmed by an experienced clinical psychologist using the Autism Diagnostic Interview-Revised (ADI-R) (Lord et al., 1994; Rutter et al., 2003) and a review of their most recent developmental and medical reports. The ADI-R is a detailed semi-structured interview of parents about their child's developmental history and autism symptoms that yield ratings for qualitative abnormalities in reciprocal social interaction (Score A), language, and communication (Score B), restricted, repetitive, and stereotyped patterns of behaviors (Score C), and abnormality of development (Score D). The scoring algorithm is similar to the diagnostic criteria of ICD-10 and DSM-IV. It is comprised of 93 items, 42 of which can be ranked into the following four scores with the respective cutoff values for diagnostic purposes: Score A- 10; Score B- verbal 8; Score C- 3; and Score D- 1. All participants in the experimental cohort of the study have results that meet the requirement the child must score above the cut-off level in each of the three domains and exhibit some abnormality in at least one area by age of 36 months, and they were classified as patients with autism according to their scores from ADI-R (see **Table 2**).

Thirty-two typically developing children and adolescents, matched for age and sex to the ASD group, formed the control sample (24 boys and 8 girls; mean ± SD [range] age = 11.4 ± 2.3 [8–16] years). They were recruited from local schools and attended regular school classes at expected grade levels. The parents confirmed in writing that their child did not have a history of any mental or neurological diagnosis.

As expected, an independent-samples *t*-test confirmed that the two groups did not differ in age: $t(54) = 0.324$, $p = 0.747$, and sex

TABLE 1 | Sample characteristics.

	ASD group (*N* = 24)	TD group (*N* = 32)
N (male/female)	24 (16/8)	32 (24/8)
Age Mean ± SD [range] in years	11.6 ± 2.4 [8–16]	11.6 ± 2.4 [8–16]
WISC-IV (Mean ± SD [range])		
VCI	81.62 ± 18.42 [45–124]	105.15 ± 11.11 [85–142]
PRI	90.00 ± 22.75 [50–136]	99.46 ± 13.61 [76–129]
WMI	86.50 ± 18.22 [59–123]	103.43 ± 10.97 [77–123]
PSI	84.87 ± 16.82 [55–139]	99.18 ± 12.52 [76–124]
FSIQ	84.04 ± 17.24 [59–122]	102.28 ± 13.30 [80–141]

TABLE 2 | ADI-R domain-specific scores.

	ASD group (*N* = 24)
ADI-R (Mean ± SD [range])	
Score A Qualitative Abnormalities in Reciprocal Social Interaction	26.16 ± 4.47 [11–30]
Score B Qualitative Abnormalities in Communication	19.66 ± 4.21[9–24]
Score C Restricted, Repetitive, and stereotyped behavior	7.12 ± 2.77 [2–12]
Score D Abnormality of Development Evident at or Before 36 Months	4.33 ± 0.96 [2–5]

$t(54) = -0.674$, $p = 0.503$. Although the groups with ASD and TD were carefully matched in terms of age and sex, matching IQ score was a challenge as we wanted to include in the study as wide as possible group of participants from the autism spectrum, that would result in different levels of intellectual functioning, and the difference in WISC score was expected: FSIQ $t(54) = -4.471$, $p < 0.05$, VCI $t(54) = -5.934$, $p < 0.05$, WMI $t(54) = -4.322$, $p < 0.05$, and PSI $t(54) = -3.652$, $p < 0.05$. There was no significant difference in mean PRI score between ASD and TD groups $t(54) = -4.471$, $p > 0.05$.

Five of the participants in each group dominantly used the left hand. All participants had normal or corrected-to-normal near and far visual acuity, measured by Rosenbaum Pocket Vision Screener and Tumbling "E" Test, respectively at 35.6 cm and 3 m. All had 1200″ stereo acuity measured by Lang stereo test and normal contrast sensitivity measured by Hamilton-Veale Contrast Sensitivity Test.

Stimuli and Procedure

The stimuli were generated by a custom software and presented on an EIZO CS230 23″ monitor with a vertical refresh rate of 60 Hz and a screen resolution of 1920 × 1080 pixels. The stimulation field had a mean display luminance of 18 cd/m^2 and a size of 22.5 × 40° (ratio 16/9). The monitor's default settings and calibration were checked and controlled by X-Rite i1 Eye-One Monitor Calibrator. Custom software written in C + + was used to generate the stimuli by an OpenGL video card and to control the experiment.

A virtual contour (the target) of Gabor patches was embedded among similar patches with random orientation in the range of ±90°. The Gabor patches were positioned on a gray background at the intersection points of an invisible hexagonal grid of 39 columns × 25 rows. In such a way, 975 Gabor elements were generated and spaced at 1.044° (**Figure 1**). The Gabor stimuli had a spatial frequency of 5.75 cpd, a standard deviation of 0.087°, and a diameter of 0.522° with elongation 1.0 from a viewing distance of 70 cm. All Gabors were displayed at 75% Michelson contrast to avoid non-linear distortion of the monitor at very low and very high intensity. The average brightness of the stimuli coincided with that of the background. In half of the trials, the target contour consisting of 12 Gabor elements with the mean orientation of 60° was presented at the middle of the screen, as shown in **Figure 1A**. In the no-noise condition, all of the Gabor elements have a 60° orientation coinciding with the contour tilt. The external noise was defined as the orientation jitter added to the contour elements in the no-noise condition. Six noise levels (determined on a base of pilot experiments) were generated by adding or subtracting 0 (no-noise condition), 10, 20, 30, 45, or 60° to the orientation of the Gabor elements forming the contour. This manipulation preserves the mean orientation of the contour at 60°, but changes the variance of the contour elements; it is approximately equal to half of the maximal orientation change. The mean orientation of the rest stimulus elements was close to 0° with a standard deviation of about 50°. In the other half of the trials (non-target condition) the target contour was replaced by randomly oriented elements. The target or non-target stimuli were presented for 200 ms.

The precise parameters of the stimulation, such as the stimulus duration and noise levels were chosen based on pilot experiments in order to find the most suitable values for obtaining perceptual performance above the guess level and below 100%. A group of children and adolescents (6–16 years old) took part in the pilot experiments. We selected the method of constant stimuli as, if we have used an adaptive procedure, we would not be able to compare the performance of the participants in identical conditions; we would obtain only one value–the threshold representing the contour degradation the observers could tolerate, but we would miss the information about the participants' sensitivity to the contour presence when no noise is added to the contour or at high noise levels.

The Yes/No procedure was used. The observers' task was to indicate "as accurate and as fast as possible" (with the emphasis on the accuracy) the target presence or absence by pressing appropriate predetermined buttons on a controller. The six noise levels of the contour Gabor elements were separated into different experimental blocks. The separation of the stimuli in blocks reduces stimulus uncertainty and gives the participants the opportunity to adjust their perceptual template to the stimulus variability. Each block included 60 randomly ordered trials: 30 trials of target condition containing the contour and 30 trials with the non-target condition without a contour. The first trial was initiated by the participant pressing any button. Each next trial was triggered by the participant's response to the previous trial. After an intertrial interval of 2000 ms the new trial started with the appearance of a blank gray screen of mean luminance with a fixation dot in the center accompanied by a warning beep. After a fore-period that varied between 500 and 1000 ms, the blank screen was replaced by target or non-target stimulus. The participants were instructed to look at the fixation dot, which coincides with the center of the target contour stimulus if it appears. Each experimental block started with six training trials: three trials contained target stimuli and three – non-target stimuli, the responses to which were disregarded.

Before the start of the first experimental block, stimuli at all noise levels with an unlimited stimulus duration were demonstrated to each observer and at least 1 training session at different noise levels was performed. Participants were given self-timed breaks between the separate blocks.

During each experimental trial, the gaze positions of the observer were recorded by the Gaze tracker Gazepoint GP3HD Desktop. The spatial accuracy of the eye tracker is 0.5–1°, and the

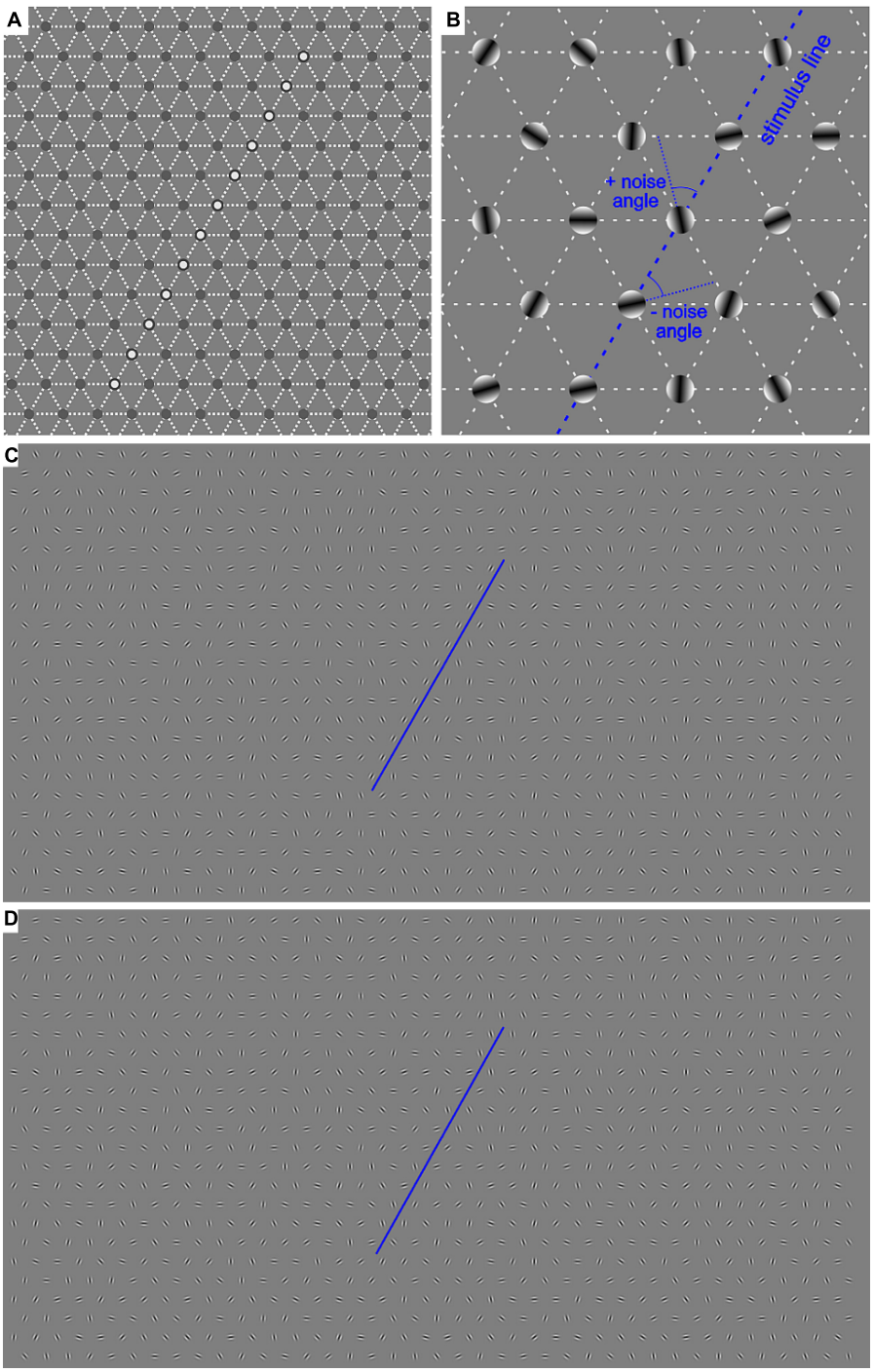

FIGURE 1 | (A) Example of the central area of the invisible hexagonal grid on which the Gabor patches were positioned. The white circles in the most central area denote the positions where the contour appeared in half of the trials. **(B)** Demonstration of the generation of the external noise added to the contour. **(C,D)** Examples of the whole screen with a contour in the no-noise condition (0°, **C**) and in 45° noise condition **(D)**. The blue lines underline the contour stimulus.

resolution was set at 150 frames per second. The calibration was done with nine points of fixation and was checked with 11 points. If the check was not of good quality, the calibration was repeated.

The participants were in a darkened room without direct sunlight. The viewing was binocular, at a distance to the screen of 70 cm. The viewing distance of 70 cm was ensured by the fixed distance between the table under the monitor and the participant's chair. The distance was verified periodically by using the gaze tracker control. Participant responses were collected via color-coded keys on a joystick controller. The responses,

including the reaction time (RT), were processed by a custom device and transmitted to a computer.

In addition, a double pass paradigm was employed to assess internal noise (Burgess and Colborne, 1988; Vilidaite et al., 2017). Experimental blocks at two noise levels: low, 10°, and high, 45°, were repeated twice (two passes) by each participant in different daily sessions. The first pass was run in a predetermined random order, followed by the second pass with an identical stimulus presentation order.

Thus, each participant performed eight blocks altogether: six blocks at the six noise levels and two additional blocks repeated at the noise levels of 10 and 45°. The blocks of different noise levels were run in random order. The additional two blocks of 10 and 45° were always run last. In order to minimize tiredness, the data was obtained in 2 or 3 sessions of 2–4 blocks of trials, measured on different days.

After the procedures were fully explained (the details of the project and a participant information sheet), the parents provided informed written consent before inclusion. Informed consent was obtained orally from each participant. The decision regarding participation in the project was entirely voluntary. Participants received a voucher as a reward for participation. A researcher emphasized to the participants that their consent could be withdrawn at any time without penalty or affecting the quality or quantity of their medical/social or educational care, or loss of benefits to which the participant was otherwise entitled. One copy of the informed consent form was kept by the participant's parents, while the other was kept by the research team. The experimental procedure was in accordance with the ethical standards of the Declaration of Helsinki and its later amendments or comparable ethical standards and was approved by the Ethics Committee of the Institute of Neurobiology, Bulgarian Academy of Sciences. All participants were cooperative and understood the task, as demonstrated by their performance in training trials.

Statistical Analyses

All analyses included in the paper were performed in the R environment (R Development Core Team, 2014).

To compare the processes of contour detection performance in the two groups with different development, we used the bayesboot package (Baath, 2018) on the proportion of correct responses and the reaction time. The analysis allowed to estimate the confidence limits of these two characteristics of the performance using the values corresponding to 2.5 and 97.5% of the posterior distributions at each noise value. The default sample size of 4000 values was used. The probability of significant differences between the two groups at each noise value was also estimated. For the reaction time, we excluded all response times that were less than 0.25 s and more than 4.0 s as outliers.

To analyze the effect of noise level and the group on proportion correct responses, we use the lme4 package (Bates et al., 2015) for fitting a generalized linear mixed model regression for the binomial family with a logit link. In the analysis, we also used the IQ scores as a continuous predictor.

Also, we evaluated the relationship between the accuracy and the consistency of the responses in the double-pass of the experimental conditions at noise values of 10° and 45°. We used the methodology of Gold et al. (2004) to evaluate the ratio of the internal to the external noise σi/σe. This ratio was estimated from the following equation:

$$p_c = m^{*}\log_{10}(p_a/100) + 100 \quad (1)$$

In Eq. 1, p_c is the percent of correct responses, p_a – the percept of agreement between the responses from the two passes of the experiment and the slope m represents the ratio of the internal to the external noise. We used the nlme package (Pinheiro et al., 2020) to evaluate the two different values of noise for the two groups and the package emmeans (Lenth, 2019)–to evaluate whether the slopes differed. As we used two different values of external noise, one low and one high, the difference in the slopes will indicate whether the internal noise is additive or stimulus-dependent (multiplicative).

To analyze the effect of the group and the added external noise on the response time, we applied a generalized linear mixed regression model using the glmmTMB package (Brooks et al., 2017). We used Gamma distribution with an "identity" link function, as suggested by Lo and Andrews (2015). We also included in the analysis the IQ scores to evaluate the potential role of the intellectual abilities on response time.

In addition, the responses were separated into four categories according to the Signal Detection Theory (Green and Swets, 1966): hit (signal present and subject says "yes"), miss (signal present and subject says "no"), false alarm (signal absent and subject says "yes"), and correct rejection (signal absent and subject says "no"). The data in the different categories were used to verify the effect of the group and noise on the average percentage of the different response types for each participant at the different noise levels.

For the eye positions of the participants, we used spatial point pattern analysis (Baddeley et al., 2016). The mean coordinates of gaze positions for each trial and their standard deviations were estimated. We considered the distribution of gaze positions as spatial point patterns. We included in the analyses only the gaze positions allocated inside the presented image (i.e., inside the screen). As a result of this choice, 10% of the data of participants with autism and 4% of the data of participants with typical development were excluded from consideration. To compare the effects of noise and the differences between the two groups with different development, we used tools from the spatstat package (Baddeley and Turner, 2005). As the point patterns were generated by the eye positions from different trials, we considered them as independent and hence, as generated by a Poisson point process. A homogeneous distribution for a Poisson point process would imply complete spatial randomness. To evaluate whether the gaze positions are evenly distributed or clustered, we used the quadrat test. We also checked whether there was a difference between the distributions of the eye positions in trials when the contour was present (signal trials) and in the trials when only noise elements were presented (noise trials). For this purpose, we marked the points in the pattern depending on the type of stimuli (signal or noise) and applied a model of inhomogeneous Poisson distribution to the data. We used a second-order polynomial to describe the intensity (the expected

density of points per unit area) of the distribution of the points as a function of their spatial coordinates. This choice implies the assumption that the gaze positions will be distributed in an elliptical region. The ppm function was used. This function is analogous to fitting a linear or generalized linear model to the point patterns.

To evaluate the contribution of the individual differences in each group on the variability of gaze positions, we used the pairdist function that gave the distance between all pairs of points in a pattern and estimated the summary statistics of the distances for different noise levels and groups. To determine whether each observer fixated the same locations on the screen for each noise level, we estimated the standard deviations of gaze positions in the repeated presentation of stimuli with the same added noise for each participant. We compared the differences in their distributions for each noise level using the bayesboot package (Baath, 2018).

RESULTS

In the present study, we recorded three types of performance characteristics: the response to the presence of contour, the response time, and the gaze of the participants. Below we present sequentially the analyses of these characteristics aiming to answer the question of whether the added external noise to a contour affects differently the detection performance of the two groups with different development.

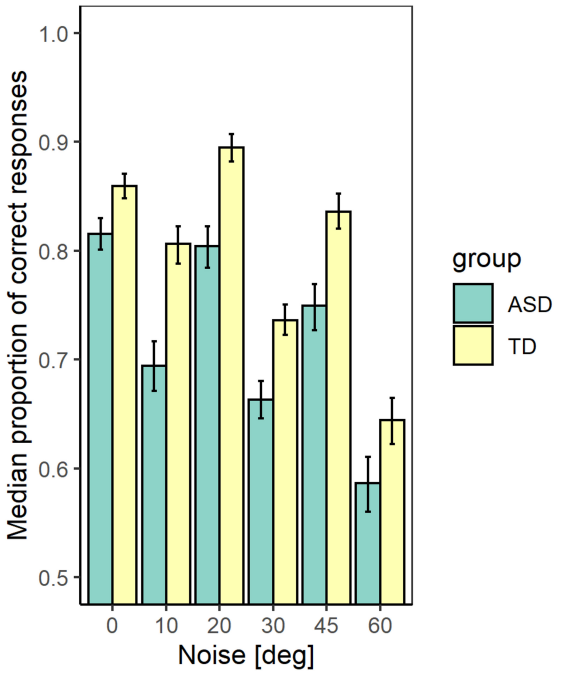

FIGURE 2 | The median values of the proportion correct responses for the two groups with different development at different noise levels. The error bars correspond to the 2.5 and 97.5% values of a bootstrap sample of size 4000.

Effect of the Added External Noise on Sensitivity

Figure 2 represents the median values of the correct responses of the participants from each group and noise level with confidence limits obtained from Bayesian bootstrap. The figure shows that at all noise levels the participants from the TD group achieved higher accuracy than the participants from the ASD group. We estimated the correlation between the mean proportion of correct responses and the IQ scores. The results show a significant positive correlation ($r(54) = 0.63$ [0.44 −0.77]; $p < 0.001$), implying that the detection performance depends on the intellectual abilities of the observers. To evaluate whether the group differences affect the performance irrespective of the intellectual abilities, we performed a generalized mixed model regression on the proportion correct responses, including as continuous predictors the noise level and the IQ scores and the group as a between-group factor. A random intercept and slope were included. The results show a significant effect of the noise level ($\chi^2(1) = 179.04$, $p < 0.001$), of the group ($\chi^2(1) = 6.94$; $p < 0.01$), and the FSIQ ($\chi^2(1) = 4.25$; $p < 0.05$). The interaction between the noise level and the group is non-significant ($\chi^2(1) = 1.77$; $p = 0.18$). The results show that the accuracy of contour detection evaluated by the proportion of correct responses decreases with the increase of the added external noise and increases with the IQ of the participants.

We verified the effect of the group and noise on the average percentage of the different response types for each participant at the different noise levels. The responses were separated into four categories according to the Signal Detection Theory (Green and Swets, 1966): hit, miss, false alarm, and correct rejection. The results show that both the group and noise have a significant effect on the number of hits, but their interaction is non-significant ($\chi^2(1) = 15.69$; $p < 0.01$ for the effect of the group; $\chi^2(1) = 117.79$; $p < 0.01$–for the noise effect, and χ^2 (1) = 1.62; $p = 0.20$–for their interaction). Only the effect of the noise is significant for the number of false alarms ($\chi^2(1) = 0.02$; $p = 0.98$ for the effect of the group; $\chi^2(1) = 70.85$; $p < 0.01$–for the noise effect, and $\chi^2(1) = 2.30$; $p = 0.13$–for their interaction) as well as for the number of correct rejections ($\chi^2(1) = 0.79$; $p = 0.37$ for the effect of the group; $\chi^2(1) = 165.51$; $p < 0.01$–for the noise effect, and $\chi^2(1) = 0.58$; $p = 0.44$–for their interaction). For the number of misses, all effects are significant ($\chi^2(1) = 14.53$; $p < 0.01$ for the effect of group; $\chi^2(1) = 165.51$; $p < 0.01$–for the noise effect, and $\chi^2(1) = 5.33$; $p < 0.05$–for their interaction). The proportion of misses is lower for the TD group at all noise levels, but it increases more strongly with the noise increase for this group than for the ASD group. Sensitivity to contour detection depends on the proportion of hits and false alarms. Hence, our data imply an inferior ability to detect the contour presence for the ASD group. The deteriorated ability of contour detection is also supported by the higher proportion of misses for this group.

Using Eq. 1 and the data of the double-pass experiment, we obtained that the ratio of the internal to external noise m is 0.82 [0.77 −0.88] and 0.86 [0.81 −0.90] for the ASD and the TD group, respectively at noise level 10° and 0.67 [0.61 −0.72], 0.74 [0.69 −0.78]-for the two groups at noise value 45°.

The values in brackets give the 2.5 and 97.5% lower and upper confidence limits. For both groups, the slopes at 10° and 45° differ significantly ($p < 0.001$). The difference in slopes between the two groups is non-significant at $p = 0.05$ for noise level 10° and differs significantly at a noise level of 45° (t-ratio = 3.58, $p = 0.04$). These results suggest that the internal noise for the two groups depends on the level of the added external noise, and the group of ASD participants is more affected by the increase in external noise. As the level of external noise for the two groups is the same, the lower slope for the ASD group implies either higher stimulus-dependent noise for this group or a suboptimal perceptual template. The non-significant difference of the slopes at the lower level of external noise suggests similar additive internal noise levels–the major limiting performance factor at low external noise levels.

Figures 3, **4** show the dependence of the percent correct responses on the proportion of agreement for the two groups for the two noise levels.

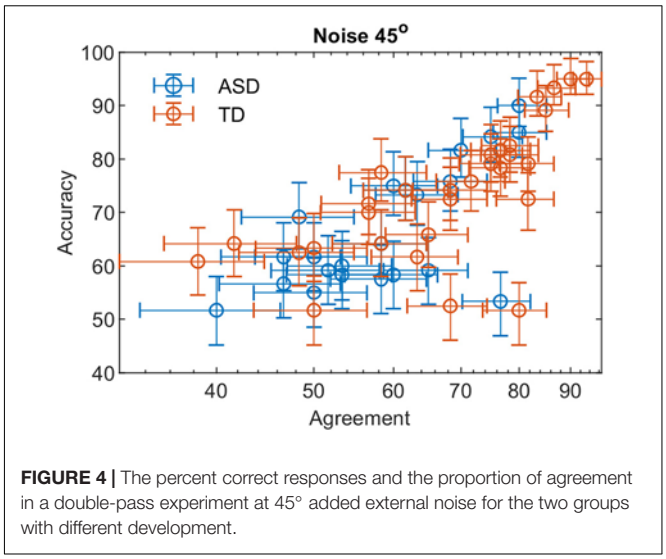

FIGURE 4 | The percent correct responses and the proportion of agreement in a double-pass experiment at 45° added external noise for the two groups with different development.

Effect of the Added External Noise on Reaction Time

Figures 5, **6** show the median values of the RT for the participants of the two groups and the variability of its values at each noise level obtained using Bayesian bootstrap. The results imply that in all conditions, the RT for the ASD group is longer than the RT of the TD group and has a higher variability. For both groups, the increase in the external noise leads to an increase in the RT and its variability. The response time reflects different cognitive processes, some of them related to decision-making, others–to the encoding of the stimulus information and the motor response's preparation and execution (Ratcliff and McKoon, 2008). For example, the response time may increase due to the observers' attempt to keep high accuracy and hence, needing more evidence before making a choice. It could also depend on the task difficulty that affects the rate of evidence accumulation. We estimated the correlation between the mean response time and the IQ scores to evaluate whether the observers' intellectual abilities affect their response time. The results show non-significant correlation between these two characteristics ($r(54) = -0.24$ [−0.47 −0.03], $p = 0.07$. This outcome suggests that the differences between the two groups observed in **Figure 5** might be related to stimulus encoding and motor response preparation processes.

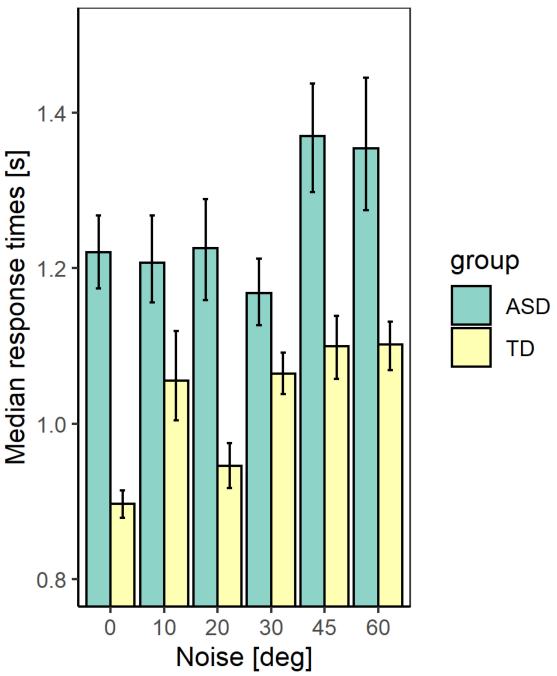

FIGURE 5 | The median values of the response time for the two groups with different development at different noise levels. The error bars correspond to the 2.5 and 97.5% values of a bootstrap sample of size 4000.

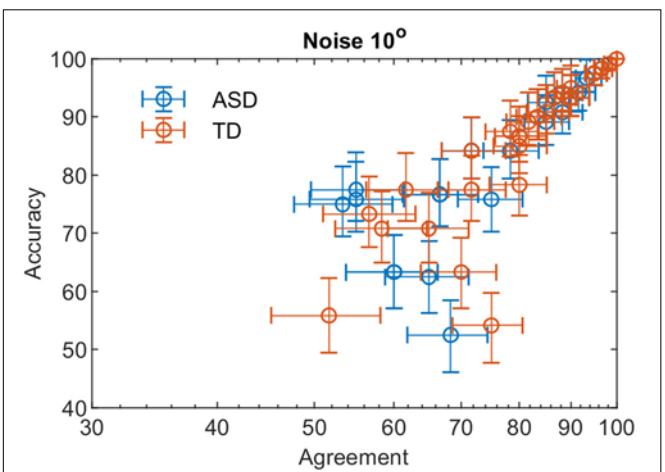

FIGURE 3 | The percent correct responses and the proportion of agreement in a double-pass experiment at 10° added external noise for the two groups with different development.

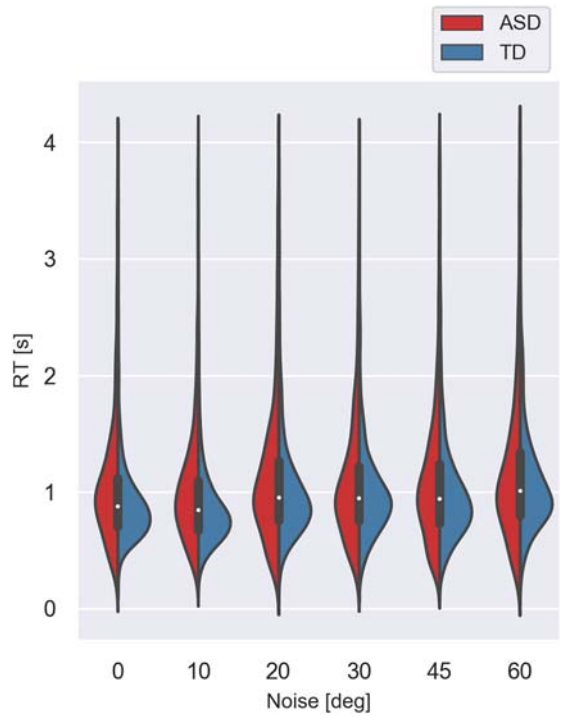

FIGURE 6 | Violin plot of the variance of response times for the two groups with different development at different noise levels. The values lower than 0.25 s and higher than 4.0 s were excluded.

We performed a generalized mixed linear model on the response time using as continuous predictors the levels of added external noise and the IQ scores and the group as a between-subjects factor. We included random slopes and intercepts to account for the individual differences. The results show significant effects of the noise level ($\chi^2(1) = 24.04$; $p < 0.001$) and the group ($\chi^2(1) = 8.11$; $p <$,01). The effect of the IQ score ($\chi^2(1) = 0.01$; $p = 0.92$) and the interaction between the noise level and the group ($\chi^2(1) = 1.69$, $p = 0.19$) are non-significant. The increase in the noise level leads to a prolongation of the response time that could reflect a change in the task's difficulty with increased noise. The outcome of the analysis, however, implies that even though the ASD group responds more slowly, the noise affects their response time similarly to the TD group.

Effects of the Added External Noise on Gaze Allocation During Stimulus Presentation

The experiment was conducted with the presentation of a fixation point located in the center of the stimulus. In the trials, when the contour was presented it always appeared in the same location. Therefore, it can be assumed that the participants will maintain a stable fixation during the stimulus duration because it was only 200 ms. For this reason, it can be expected that there will be no difference in the distribution of the gaze positions between the different experimental blocks, corresponding to different levels of added noise to the contour, as well as between the two groups with different development. However, if the fixation stability differed between the two groups, a difference between the distributions of gaze positions might occur. Still, no difference between the blocks with different noise levels is expected. A third potential scenario is that the participants direct in advance their gaze to the parts of the image that they expect to carry the most relevant information about a contour's presence. The redirection of the gaze is carried out during the presentation of the fixation point. In this scenario, a different distribution of gaze positions may be expected depending on the participants' group and the amount of noise added to the contour.

First, we evaluated whether the gaze allocations were evenly distributed or are clustered. The analysis showed that for all experimental conditions and all groups studied, the eye positions were clustered, and their distributions are inhomogeneous (quadrat count test, $\chi^2(24) = 21630$ and 22164 for children with ASD for the signal and noise trials, respectively, and 149924 and 151170 for children with TD in these conditions, $p < 0.001$). The graphical comparison of the envelope of Ripley's K-function showed that the gaze positions form point patterns that are not only inhomogeneous but also that for the autistic group the distance between the points is greater than expected based on the estimated intensity function for non-homogeneous patterns.

We tested whether the distributions of the gaze positions for the signal and noise trials differ. We fitted an inhomogeneous Poisson point process model on the intensity of the point patterns as a second-order polynomial function of their spatial coordinates separately for each group. This analysis showed that for both groups, the distribution of eye positions did not depend on the presence or absence of a contour ($\chi^2(1) = 0.73$; $p = 0.39$ for children with ASD and 0.23; $p = 0.69$ for children with TD). The lack of difference between the signal and noise trials is expected as the observers could not know the type of the presented stimulus in advance. It may be due to maintaining constant fixation or to the use of the "history" of the presented stimuli to predict the most informative parts of the images in determining the presence of a contour. To distinguish between these two hypotheses, we created a hyperframe, i.e., a data frame that contains objects of any kind. We included in the hyperframe the point patterns obtained for stimuli with different levels of added noise for the two groups of participants and tested the effect of these factors on the gaze allocation. Here again, we assumed that the intensity of the point patterns depends on the spatial coordinates of the points as a second-order polynomial. The point patterns were considered as samples from inhomogeneous Poisson distribution. The results show a significant effect of the noise level ($\chi^2(5) = 967.54$; $p < 0.001$) and of the group ($\chi^2(1) = 2321.30$; $p < 0.0001$), as well as a significant interaction between the level of the added noise and the group ($\chi^2(5) = 206.82$; $p < 0.001$). There are also significant effects of the spatial coordinates ($\chi^2(5) = 13409.29$; $p < 0.001$ for the combined effect of the 5 elements of the second-order polynomial: x, y, x^2, y^2, and x*y), of the interaction between the noise level and the

spatial coordinates ($\chi^2(25) = 1559.46$; $p < 0.001$), and of the interaction between the group and the spatial coordinates ($\chi^2(5) = 2539.59$; $p < 0.001$). The triple interaction between the spatial coordinates, the noise level, and the group is also significant ($\chi^2(25) = 2123.80$; $p < 0.001$). The effect of noise on the distribution of gaze positions implies that the participant might be using the "history" of stimulus presentation to locate the contour.

The comparison of the distribution of the pattern intensity indicates that the position of the pattern in horizontal (x) and vertical direction (y) differs at all noise levels except at noise level 0° (no added noise). The intensity of the gaze point patterns does not differ significantly in vertical direction also at noise level 45°. At all other noise levels, the distribution of gaze positions differed in x^2, x*y, and y^2 implying different elongation and spread of the gaze positions. Whereas the effect of the group on the pattern intensity might be due to the higher number of gaze records for the typical children in the screen area, the interaction between the group, the spatial coordinates, and the noise level implies that the children from the two groups allocate their gaze to different portions of the image at the different noise levels.

We also calculated the variance ellipses of the gaze positions at different noise levels for the two groups. We estimated first whether the variance ellipses could be regarded as elongated, i.e., whether there is a significant difference between the maximal and the minimal variance of the distributions. In all cases, the *F*-test for variance comparison suggests that the distributions could be regarded as elongated (F = 2.18, 2.25, 2.36, 3.26, 2.66, 3.33–for the ASD group and 2.78, 2.61, 1.99, 3.18, 1.66, 2.69–for the TD group; $p < 0.01$). We also compared separately the maximal and the minimal variance of the gaze positions for each noise level for the two groups. For all noise levels, the maximum variance was greater for children with ASD than for the TD group (*F*-test: 1.64, 3.42, 3.02, 1.75, 2.32, 4.29 for noises from 0 to 60°, $p < 0.01$). Also, the minimal variance of the distributions for the children with ASD significantly exceeded those for children with TD (*F*-test: 2.09, 3.96, 2.55, 1.71, 1.44, 3.47; $p < 0.01$). These results suggest greater variability of eye positions for children with ASD that may be due to decreased fixation stability or larger individual differences in the selection of the most informative sections of images at different noise levels.

To discriminate between these two possibilities, we calculated the mean gaze positions for each participant and created a new point pattern using the two groups as marks. We estimated the distance between all pairs of points for each group. If the mean gaze positions of the different participants are closer, the distance between them will be smaller than if they are more dispersed. Therefore, the distribution of the distances between the mean gaze positions of any two group members could be used to measure the individual differences in this group. The median value obtained for the distances between the mean gaze positions of each pair of participants in the ASD group is 0.203 [0.110 −0.339], and it exceeds the median value of 0.124[0.059 −0.0240] for the TD group significantly (the values in brackets are for the first and the third quartiles). These results suggest more considerable individual differences in mean gaze positions of the ASD group than in the TD group.

These results may imply that the effect of noise has a different impact in the ASD group, increasing the dispersion of the mean gaze positions or that at each level of noise, the individual differences of the eye positions for this group are larger than for the TD group. The comparison of the median values of the paired distances for each noise value implies higher variability for the ASD group.

We also use Bayesian bootstrap to evaluate the differences in the variability of the gaze positions of the members of each group at a different noise level. This measure would indicate whether each participant fixated the same screen position in the block of trials with the same external noise. The results suggest a higher variability of gaze positions in the ASD group than in the TD group for noise levels of 20°, 45°, and 60°. The higher variability of gaze positions for the ASD group in comparison to the TD group at 45° and the non-significant difference at 10° noise level may be interpreted as an indication that the higher ratio between the internal and the external noise obtained from the double-pass experiment may be caused by the higher variability of the gaze positions in the ASD group. Since the variability is stimulus-dependent, it might suggest an unstable, noisy, or suboptimal perceptual template of the ASD group. The improper template would lead either to the omission of important stimulus information or the inclusion of irrelevant features and hence, to reduced ability to filter external noise.

Figure 7 shows a histogram of the gaze positions on a 31 × 31 grid in the screen window for each group at each noise value.

We also estimated the correlation between the area of gaze positions and the proportion of correct responses and the response time to test whether the fixation instability could cause a deterioration in task performance. The correlation coefficients are significant ($r(54) = -0.45$ [−0.66 to −0.18], $p < 0.01$ for the proportion of correct responses and 0.33 [0.08 − 0.55], $p < 0.05$ for the response time). The values in brackets show the 95% confidence intervals. The significant correlations imply that when the gaze is spread over a larger area, the observers are less accurate in detecting the contour and need more time to make a choice.

To test whether the intellectual abilities affect the spread of the gaze distributions, we estimated the correlation between the area of gaze positions and the IQ score. The significant negative correlation of $r(54) = -0.34$ [−0.55 to −0.09], $p < 0.05$, indicates that children with lower intellectual abilities have more dispersed gaze positions.

In summary, the analyses of the gaze positions show significant differences between the two groups with different development depending on the noise level added to the contour embedded in the background noise. These findings could be interpreted as an indication that the two groups have a different choice of which portion of the image is more informative for the presence of a contour and that this choice depends on the level of added noise. Also, the gaze positions of the children with ASD are more dispersed, implying greater individual differences and greater instability in fixations.

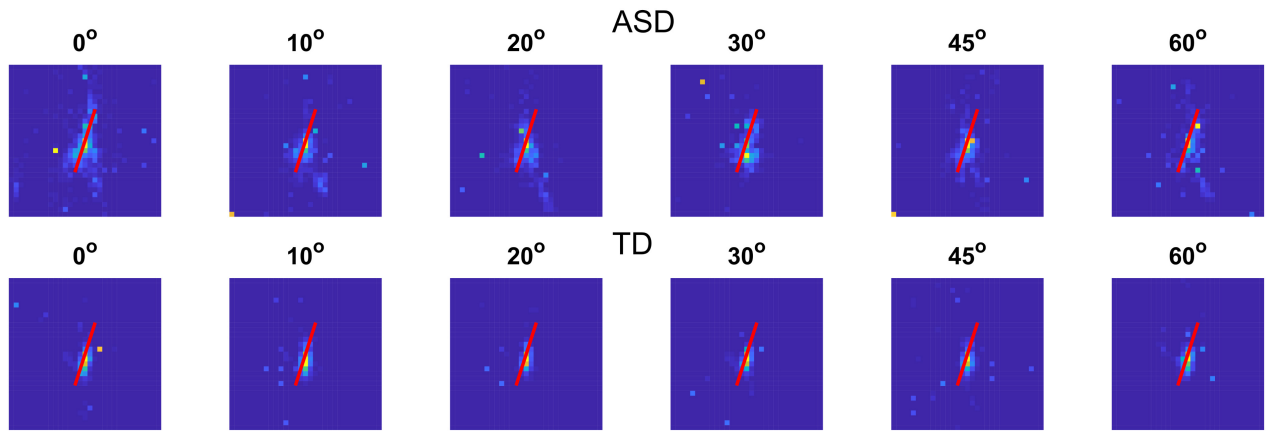

FIGURE 7 | Histogram of the gaze locations on a 31 × 31 grid for the two groups with different development at different noise levels. Gaze positions outside of the screen were excluded. The red line shows the contour position.

DISCUSSION

The results obtained in the present study showed atypical contour integration processing in autism, probably due to difficulties in rejecting background noise and integrating the elements of a jagged contour. The responses of the group with ASD were less accurate and significantly slower compared to the control group, even in the no-noise condition. In line with the psychophysical data, the eye-tracking results showed a larger gaze allocation area in the ASD. Our findings also indicate that the response time changes with the level of added external noise similarly for the two groups with different development remaining longer for the ASD group. The dependence of the response time on the stimulus characteristics suggests that either the rate of evidence accumulation (the component of response time that depends on task difficulty) or the time needed to encode the stimulus characteristics increases with the noise level increase. It also implies that the response time prolongation in the ASD group may be predominantly due to factors related to the motor response preparation and execution. The external noise added to the contour had a larger effect on gaze positions of ASD participants inducing larger dispersion of the mean gaze positions and higher variability in the ASD group. The significant correlation between the area of the gaze positions of each participant and the mean proportion of correct responses and the mean response time implies that the area of gaze positions affects children's ability to detect the contour. The comparison of the agreement and accuracy of the responses in the double-pass experiment showed that the participants with ASD are more affected by the increase of the external noise. It turned out that the internal noise depends on the level of the added external noise: the difference between the two groups was non-significant at the low external noise and significant at the high external noise.

There are many differences between our research and the previous studies investigating contour integration in ASD individuals, like the experimental procedure, the sample size, the choice, and the characteristics of the participants. We will first discuss the potential effect of these differences before focusing on our study's main distinguishable feature: contour position and noise manipulation.

Effect of the Experimental Procedure and Contour Characteristics

Since the pioneering work of Field et al. (1993) in contour integration studies, including those that involve participants with ASD, the forced-choice procedure is the most explored approach, being a temporal two-interval forced-choice (e.g., Jachim et al., 2015) or spatial four-alternative forced-choice (e.g., Del Viva et al., 2006). Although we used open contours and a detection task, our data are in line with studies that show diminished contour integration in the ASD group (Pei et al., 2009; Evers et al., 2014; Jachim et al., 2015). In fact, the performance of ASD participants was not diminished in the open contour integration task in Jachim et al. (2015), probably because of the small group size as suggested by the authors. However, the benefit from the closed contours was reduced in the ASD group, which led the authors to conclude weaker contour integration in adults with ASD. Gowen et al. (2020) replicated the findings of Jachim et al. (2015) about the reduced closure effect in autistic individuals with a new larger group of participants with ASD. However, in contrast to the first study, the result from the newest study (Gowen et al., 2020) found differences for the open stimulus between groups with ASD and TD: the perceptual performance was even better for the autistic than for the non-autistic group.

Probably, the number of contour elements could reduce the contour integration ability of our ASD group. The number of contour elements used in the present study is lower (12 Gabor patches) in contrast to the many more elements, 20 and 35, that constructed the contours, respectively in studies of Jachim et al. (2015) and Gowen et al. (2020). More elements could enhance autistic performance as the comparison of the results from the works mentioned above shows. This assumption is also supported

by neurophysiological results that contour detectability improved with the increase in the number of collinear line elements (Li and Gilbert, 2002).

Effect of Sample Size and Individual Characteristics

The groups of participants with ASD in Jachim et al. (2015) and Gowen et al. (2020) were smaller, more compact, and homogenous (the samples included only participants with a diagnosis of Asperger's syndrome) than our group with ASD. Moreover, in contrast to our group with ASD, participants in their studies were adults, 18–42 years old. This could also influence the results because there is a prolonged development for contour processing, as suggested by Taylor et al. (2014). The sample size and the age range of participants in the study of Evers et al. (2014) have several similarities with our cohort. The age range of the children and adolescents was similar to ours: 10–17 years old compared to 8–16 years old in our study. They used ADOS to confirm the diagnosis and to measure the severity of the ASD symptoms. The participants' scores ranged from 2 to 9 or from 3 to 10 (severity scores), 4–5 scores (ASD-classification), and 6–10 scores (receiving an autism classification) (Gotham et al., 2009). In such a way, some children are outside of the ranges for ASD, again raising the question of severity measurement. The group with ASD in Evers et al. (2014) was as large as our group and they found diminished identification performance of the children with ASD (see the severity score range). However, the task in their study was more complicated and it is not clear if the results are due to the larger group or a more complex task.

Because ASD is a complex, pervasive, highly heterogeneous condition with multiple subtypes and developmental trajectories, the size of the group and the choice of participants included in the study could also influence the results obtained. In order to encompass as many as possible cases from the autism spectrum, we tried to include a large sample representative for the heterogeneity of the disorder, where participants with ASD were not excluded based on their cognitive level functioning as could be seen in **Table 1**. The FSIQ score ranged from 59 to 122 in the ASD group and was significantly different from the FSIQ score of the TD group. Other scores, VCI, WMI, and PSI, also differed significantly for both groups of participants. Curiously, the PRI score, which could be presumably the most related to the performance of the visual task in our study, did not differ significantly between the ASD and TD groups. In addition to the IQ scores, Gowen et al. (2020) discussed that the autism severity could be connected to contour integration results through variability in the integrity of lateral interactions (as suggested by Dickinson et al., 2018). Using a steady-state visual evoked potential paradigm, they found that greater ASD symptom severity, assessed as an increased ADOS score, is associated with increased short-range lateral inhibition. The severity of the autistic disorder is a complicated topic, and the accurate assessment is still a challenge. DSM-5 includes a severity marker based on the degrees of impairment in the domains of social communication and restricted and repetitive behaviors. Although qualitative differences between impairment levels are described in the classification (DSM-5; American Psychiatric Association [APA], 2013), quantitative methods for differentiating between these levels are still a problem. Levels of impairment in children with ASD are usually associated with language delay, cognitive functioning, or behavioral issues such as aggression. Although these factors are important in the overall adaptive functioning, they are not core features of the autism spectrum. Notwithstanding that ADI-R could not assess directly the severity of symptoms, it should be noted that the mean group results in the present study in each of the three domains are high, and all participants in the experimental cohort are classified as patients with autism according to their scores from ADI-R since all they have results above the cut-off level in each of the three domains and exhibit some abnormality in at least one area by the age of 36 months.

Our results showed a relationship between the spread of the gaze positions, the proportion of correct responses, the response time, and the IQ scores. IQ scores also affect the accuracy of the task performance. The detection performance depended on the group differences in addition to the effect of the intellectual functioning of the participants from both groups. These results do not, however, represent the complicated picture for an individual. As an illustration of the relationship between the IQ scores, symptom severity, and contour integration performance, we decided to compare the data of the participants of the same age from our group with ASD. Moreover, this will allow capturing what Hodkinson and Hodkinson call "lived reality" (Hodkinson and Hodkinson, 2001) and to avoid the group results to absorb the individual ones. We found three male participants (Subjs. 2, 3, and 14) at approximately the same age (13.7, 14, and 13.7 years old). **Figure 8** presents the results of the three participants: the proportion of correct responses (**Figure 8A**), ADI-R- Diagnostic Algorithm Score Summary and Cutoffs (**Figure 8B**) and VCI, PRI, WMI, and PSI scores assessment by WISC IV (**Figure 8C**). The figure clearly shows that the much better perceptual performance of Subj. 2 compared to Subjs. 3 and 14 could not be explained by the potential difference in any of the scores from the psychological assessments. Moreover, **Figure 8A** shows different individual dependencies of the proportion of correct responses on the external noise level. Noise increase has the strongest effect on the results of Subj. 2 despite his best results in the no-noise condition. This observation implies more complicated relationships between all of the discussed factors that need to be elucidated in further research. It also implies that the performance at a low or no-noise condition that is limited predominantly by additive internal noise cannot predict the performance at high noise levels that is limited by the ability of noise filtering and the efficiency of stimulus information exploration.

Role of Internal Noise and Perceptual Efficiency in ASD

Several studies reported results that were interpreted as evidence against theories of reduced global perception in autism. Besides the already mentioned works of Del Viva et al. (2006) and Kemner et al. (2007), Gowen et al. (2020)

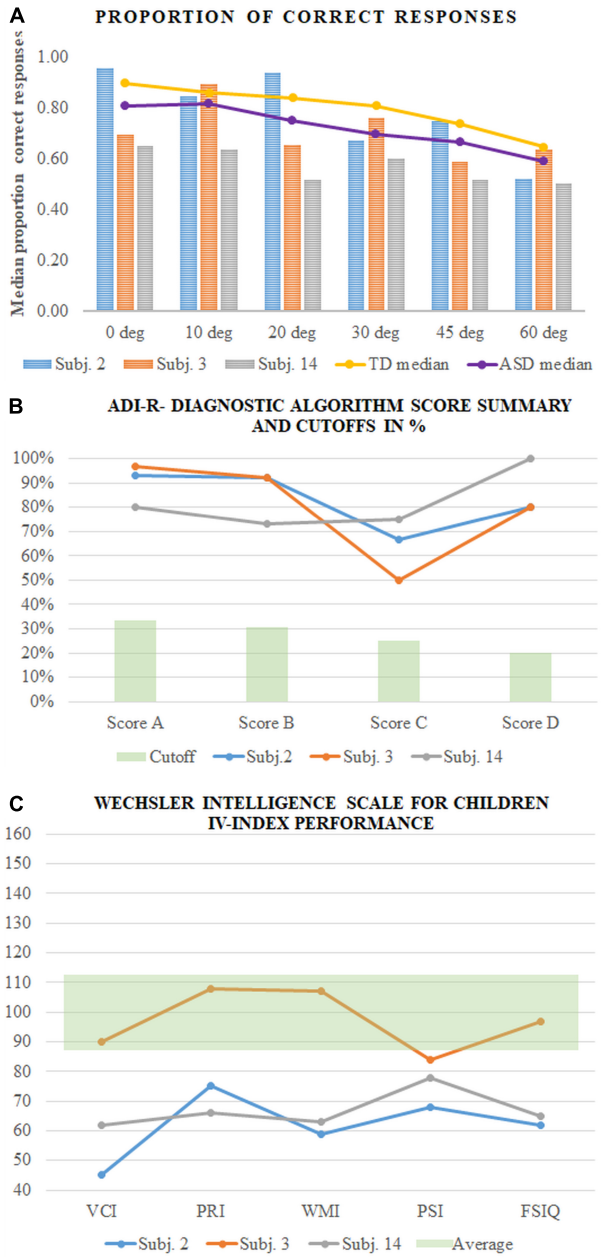

FIGURE 8 | Results of the three participants (Subjs. 2, 3, and 14 in our sample). **(A)** The proportion of correct responses of each participant compared to the medians of the group with ASD and TD; **(B)** ADI-R-Diagnostic Algorithm Score Summary and Cutoffs in%; **(C)** Wechsler Intelligence Scale for Children IV-index performance.

also found similar contour integration performance with closed, simple shapes. In line with these findings, Zaidel et al. (2015) reported intact global and multisensory integration in ASD. At the same time, the authors found a specific sensitivity to dynamic visual noise in the participants with ASD. These results were interpreted as evidence against theories of reduced global perception in autism. Zaidel et al. (2015) assumed that increased sensitivity to noise rather than diminished integration ability is a distinguishing feature of ASD.

Dinstein et al. (2015) suggested that increased neural noise in sensory and motor systems may explain why individuals with ASD suffer from different problems that affect multiple aspects of day-to-day functioning: balance problems, motor clumsiness, atypical visual perception, and abnormally large behavioral variability in trial-to-trial reaction times, eye saccade accuracy, reaching movement accuracy, and pitch of voice during speech. Moreover, neural noise theory (Simmons et al., 2007) proposes that neural noise accounts for the complex pattern of enhancements and impairment in the ASD population (see also Simmons et al., 2008, 2009).

Park et al. (2017) tried to estimate quantitatively different sources of noise that limit perceptual processing in ASD. The authors applied an equivalent noise paradigm and modeled the individual visual orientation discrimination at variable levels of external noise. It was found that the high internal noise, as well as poor external noise filtering, restricts visual processing in ASD. However, the severity of ASD symptoms correlated significantly only with internal noise estimates.

The results from the double-pass experiment in our study imply either a higher stimulus-dependent noise for the group with ASD or a suboptimal perceptual template. The non-significant difference between the groups at the lower level of external noise suggests similar additive internal noise.

In our study, the contour and the noise elements were at the intersections of regular hexagonal grid lines. The centroid positions of all Gabor elements (noise and contour) were perfectly aligned. Hence the contour detection could not be based on the positional information, but only on differences in the mean element orientation and its variability along the grid lines. The mean orientation of the contour was fixed at 60°, and we varied the orientation variance of the elements forming the contour. The perceptual organization cues that could help to segregate the contour are good continuation and similarity, and it is shown in previous studies (Avraam et al., 2019) that ASD participants could use typical perceptual organization cues. However, good continuation is an effective cue only at low added noise. The similarity could be determined either by the orientation or the variance in the contour elements' orientation. Hence, it is quite possible that the participants in our study changed their strategy depending on the external noise added to the contour. The change in strategy most probably depends on the sensitivity to the added external noise. We observed differences between the proportion of correct responses and the response consistency at low and high noise levels. One interpretation of this difference could be a change in strategy. The different spread of the gaze allocations at low and high noise could also indicate a noisier and unstable perceptual template at high noise levels. This finding raises the question of what information could the eye position measurements in our study provide.

The stimulus duration in our study is relatively short, and eye movements during the stimulus presentation could not affect the contour detection. However, we have a variable and relatively long fore-period before the stimulus appears. Our results showed significant differences in the spread of gaze positions between the two groups with different development. This finding indicates greater fixation instability for the ASD group. Few data exist on fixation stability in the age group used in the study and even less– for the group of children with ASD (Sumner et al., 2020). The study of Sumner et al. (2020) indicates that the ASD group keeps fixation for shorter times and has more intruding saccades than the TD group. However, differences between the ASD and TD groups disappear when the motor skills of the participants are taken into account. Our data provide additional knowledge about gaze characteristics of children and adolescents with ASD. Our results about the atypically larger gaze area in ASD are in line with previous results about abnormal eye control (e.g., Takarae et al., 2007) and could contribute to a better understanding of the deteriorated results on contour integration in the ASD group. They could also be considered as an indication of higher positional uncertainty in the group with ASD as compared to the TD group.

The different spread of the gaze positions in our study, however, is also related to the stimulus characteristics. It varies with the added external noise, suggesting that the gaze allocation is related to the external noise level. Due to the block stimulus presentation, the results may be interpreted as indicating that at high noise levels, the observers have difficulty determining the most informative parts of the stimulus, i.e., to have a proper perceptual template that will allow filtering the background noise and effectively using useful stimulus characteristics. The participants from both the ASD and TD groups could probably use the "history" of the presented stimuli to predict the most informative parts of the images about the contour presence, reflected in the lack of effect of the contour presence. The results of more dispersed gaze positions in the ASD group together with the stronger effect of the noise level are consistent with the assumption that individuals with ASD possess a stronger reliance on incoming sensory information and less use of prior knowledge about the world referred to as an attenuated Bayesian prior (Pellicano and Burr, 2012; Zaidel et al., 2015).

In conclusion, the results of the present study showed diminished contour integration ability in ASD, as the data were obtained from a sample that is representative of the disorder's heterogeneity. The proportion of correct responses for the contour detection was lower while the proportion of misses was higher, and the time to respond was prolonged in the ASD group at all noise levels. These results could indicate difficulties for the ASD group to integrate the elements of a jagged contour. The deviation of the individual elements from the contour path, even at the highest noise level, is in the critical limits of the associate field if used to represent contour goodness (Field et al., 1993). However, the maximum path angle that could be detected depends on the background elements' statistics (Watt et al., 2008). The deteriorated performance of the participants with ASD might be due to their inability to distinguish the target from the background noise. The comparison of the accuracy and agreement between the responses in the double-pass experiment showed that the performance of the participants with ASD is more affected by the external noise increase whilst the results of both groups were similar when external noise was low. The results obtained suggest reduced efficiency to use the available stimulus information of the participants with ASD. Also, the gaze positions of the ASD group were dispersed over an atypically large area. These findings imply lower efficiency in using stimulus information and higher positional uncertainty in the ASD group that could be caused by unstable fixation and poorer noise filtering.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Ethics Committee of the Institute of Neurobiology, Bulgarian Academy of Sciences. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

MM, NB, and TT contributed to the study conception and design. All authors performed the material preparation, data collection, and analysis, wrote first draft of the manuscript, commented on previous versions of the manuscript, read and approved the final manuscript.

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ADOS-Eye-Tracking: The Archimedean Point of View and Its Absence in Autism Spectrum Conditions

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Face perception and emotion categorization are widely investigated under laboratory conditions that are devoid of real social interaction. Using mobile eye-tracking glasses in a standardized diagnostic setting while applying the Autism Diagnostic Observation Schedule (ADOS-2), we had the opportunity to record gaze behavior of children and adolescents with and without Autism Spectrum Conditions (ASCs) during social interaction. The objective was to investigate differences in eye-gaze behavior between three groups of children and adolescents either (1) with ASC or (2) with unconfirmed diagnosis of ASC or (3) with neurotypical development (NTD) during social interaction with an adult interviewer in a diagnostic standard situation using the ADOS-2. In a case control study, we used mobile eye-tracking glasses in an ecologically valid and highly standardized diagnostic interview to investigate suspected cases of ASC. After completion of the ASC diagnostic gold standard including the ADOS-2, the participants were assigned to two groups based on their diagnosis (ASC vs. non-ASC) and compared with a matched group of neurotypically developed controls. The primary outcome measure is the percentage of total dwell times assessed for different areas of interest (AOI) with regard to the face and body of a diagnostic interviewer and the surrounding space. Overall, 65 children and adolescents within an age range of 8.3–17.9 years were included in the study. The data revealed significant group differences, especially in the central-face area. Previous investigations under laboratory conditions gave preferential attention to the eye region during face perception to describe differences between ASC and NTD. In this study – using an ecologically valid setting within a standard diagnostic procedure – the results indicate that neurotypically developed controls seem to process faces and facial expressions in a holistic manner originating from the central-face region. Conversely, participants on the Autism Spectrum (tAS) seem to avoid the central-face region and show unsystematic gaze behavior, not using the preferred landing position in the central-face region as the Archimedean point of face perception. This study uses a new approach, and it will be important to replicate these preliminary findings in future research.

Keywords: autism spectrum disorder, eye-tracking, autism diagnostic observation schedule, social cognition, social schemas, online social cognition, face perception, social interaction

INTRODUCTION

In the scope of social communication, the human face is one of the prime sources for relevant nonverbal information and an effective key instrument, producing information in a dynamic and highly efficient manner.

Within two gaze fixations, we are able to recognize a face (Hsiao and Cottrell, 2008); only a few fixations later, we can draw conclusions about gender, age, identity, ethnicity, attractiveness, health, and particularly about the emotional state of a human counterpart (Jack and Schyns, 2015). Even minute movements of unconscious facial mimicry can affect the process and development of a social interaction (Dalton et al., 2010). It follows that many researchers metaphorically speak of empathy, mimicry, and social gaze as glue for social communication (Lakin et al., 2003; Baron-Cohen and Wheelwright, 2004; Kuzmanovic et al., 2009), and the face can be considered as the focal point of direct social interaction.

People with Autism Spectrum Conditions (ASCs) show a wide range of clinical characteristics, but difficulties in social interaction and nonverbal communication are considered as core challenges for people on the Autism Spectrum (tAS). Several groundbreaking eye-tracking studies have illustrated that individuals on tAS show reduced attention to salient social stimuli, especially in the eye region (Klin et al., 2002; Jones et al., 2008; Jones and Klin, 2013). These studies, however, are all investigations conducted under laboratory conditions in which the stimulus material was detached from the participant and presented *via* screen.

What is lacking in this type of stimulus presentation is the interactive aspect of social communication in the real world (Foulsham, 2020). Pictures, comics, photographs, and video sequences of social content are passive and self-contained; in most cases, the problem definition focuses on a specific task, which the participant has to fulfill as an (passive) observer, not as an (participating, active) interactor. It is, therefore, a form of studying "offline" social cognition (Schilbach, 2014) with high internal validity, but information on the context, functioning, and processing of the rules of "online" social interaction remains poor.

Interpersonal social interaction is distinguished by a permanent exchange of social signs that are simultaneous and time constrained using limited cognitive resources and bounded rationality (Simon, 1956). In addition to the verbally mediated content, one has to perceive and categorize paraverbal modulations, body posture, gesture, and especially facial expressions such that the given response meets the expectations of the counterpart.

In order to reduce the contingency and complexity of such a social situation and to allow for context-adequate communication, the expectations of expectations (Luhmann, 1987) of the interactors have to be coupled with social schemas and scripts (Bartlett, 1932; Schank and Abelson, 1977; Augoustinos and Walker, 1995; Schaller and Rauh, 2017; Schaller et al., 2019).

As such, real-time social interaction with natural human agents in a specific contextual framework places very different demands on participants than a purely observational offline task.

Looking now at available meta-analyses concerning eye-tracking in ASC, it becomes clear that studies of autistic children and adolescents as well as those of adults on tAS show significantly reduced gaze-fixation to the eye-region of faces. A closer look at the methodology of the included studies reveals that all are based on an offline social cognition design, even those that have been specified as interactive. In this context, "interactive" is described as any static or dynamic image involving at least two human or animated figures that are posed in a possible state of interaction that has to be observed by the participant (Papagiannopoulou et al., 2014; Frazier et al., 2017).

Other survey articles, however, make clear that results found by using offline cognition are not consistent according to the hypothesis that individuals on tAS show reduced fixation of the eye-region (Guillon et al., 2014).

In their eye-tracking study, Chevallier et al. (2015) show that the ecological relevance of social stimuli is an important factor to measure social attention and motivation in ASC. Therefore, they use an interactive task. The interaction, however, is that of characters shown in a video and by no means an interaction between social stimulus and participant. On the other hand there is current evidence showing that individuals on tAS spend less time to social stimuli especially in complex social situations with more than one person (Chita-Tegmark, 2016). These examples illustrate the big heterogeneity of the methodological positions in eye-tracking studies concerning social cognition in ASC.

From this, the following question arises, to what extent do the demands of complex social cognition alter gaze behavior if the given task requires an individual to be an interactor in an ecologically valid social situation instead of just a passive observer in a detached offline task.

To answer this research question, we focused on the "Conversation and Reporting" activity within the Autism Diagnostic Observation Schedule, 2nd edition (ADOS-2, Hus and Lord, 2014) and applied a mobile eye tracking system during a 10-min sequence of social interaction in order to assess and compare gaze behavior of people with suspected diagnoses of ASC with neurotypically developed controls.

The hypothesis of this study states that – in an ecologically valid, socially dynamic situation – participants on tAS will show different proportions of total dwell times in areas of interests (AOIs) concerning the face of the interviewer when compared (a) to patients with other psychiatric diagnoses and (b) to neurotypically developed controls.

MATERIALS AND METHODS

Participants

Study participants were recruited from the population of referrals with suspected ASC from the outpatient clinic of the Department of Child and Adolescent Psychiatry, Psychotherapy and Psychosomatics of the Medical Center of the University of Freiburg within the time period from February 2014 to February 2016. In total, there were 290 children and adolescents with

initial suspicion of ASC that could be tested with ADOS-2 Module 3 or 4 as part of the gold standard diagnostics for ASC.

Inclusion criteria for participation in the study were the following: age range from 8.0 to less than 18.0 years; IQ ≥ 70; full command of the German language; and parental consent.

The list of exclusion criteria consisted of (i) vision defects that required visual acuity correction devices (>±1.5 dpt; wearing glasses is not possible in combination with the mobile eye-tracking device) and (ii) patients with severe ADHD symptoms, which could not be completely controlled by medication (high risk of invalid mobile eye-tracking recordings). Within the control group, children and adolescents with values in the clinical range for the Social Responsiveness Scale (SRS; total raw score cut-off ≥75; Bölte and Poustka, 2008) and the Child Behavior Checklist (CBCL/4–18; T-score >63 on Internalizing, Externalizing, and Total Scales; Greenbaum et al., 2004) were also excluded from further analyses.

Although only few eye-tracking studies with individuals on tAS report large effect sizes, it was clear from the beginning that we could not achieve a sample size that would have been sufficient to reveal medium effects. In order to be able to detect at least large effects between the clinical samples and the control group (power = 0.80 and alpha = 0.05), power calculations indicated n = 20 per sample and hence a total sample size of 60 children (as computed by G*Power, version 3.1.3; Faul et al., 2007).

Information on this study was provided to the parents or caregivers and the children themselves before their voluntary participation *via* a written information letter as well as a verbal description. Prior to a child's participation, the parent or caregiver was required to sign a written informed consent form.

Initially, 63 children and adolescents were recruited for the study. Some were later excluded from further analyses (see **Figure 1** for the flow of participants).

In the end, the data of 45 participants with suspected ASC could be included in the statistical analysis. In addition, a control group of neurotypically developed children and adolescents (neurotypical development, NTD; n = 20) was recruited and matched by age, IQ, and gender. In sum, the total sample consisted of N = 65 children and adolescents.

All participants were tested with ADOS-2 Module 3 or 4 (Hus and Lord, 2014), depending on age. It should be noted that none of the participants had been diagnosed with ASC prior to the study.

Accompanying Instruments

Autism Diagnostic Observation Schedule-2

The "Autism Diagnostic Observation Schedule-Generic" (ADOS-2; Hus and Lord, 2014) is a semi-structured, well-validated observational assessment. It consists of five modules: Toddler Module (pre-verbal/single words; 12–30 months old), Module 1 (pre-verbal/single words; 31 months and older), Module 2 (phrase speech), Module 3 (fluent speech; child/adolescent), and Module 4 (fluent speech; adolescent/adult). Each module consists of 11–15 parts called as "activities." In the present study, only Module 3 and Module 4 were administered; both modules incorporate the activity *Conversation and Reporting*, during which relevant eye-tracking data was registered.

Each ADOS-2 module has its own classification algorithm that is based on three components with two cut-offs each: (1) scale *Communication Total*, (2) scale *Social Interaction Total*, and (3) combined score of *Communication + Social Interaction Total*. According to the attained cut-offs, it defines three classifications: (a) *autism* or (b) *autism spectrum* or (c) *non-spectrum*. One relevant item is *B1. Unusual eye contact* judged by the diagnostician across all activities. This item has only two dichotomous values: 0 = *appropriate gaze* or 2 = *purely modulated eye contact*.

Concerning the psychometric properties of the ADOS-2 (or its precursors), there exist many studies since its development in the 1980s. For the German version of ADOS, Bölte and Poustka (2004) report the following information: the interrater and retest reliability were shown both at the level of diagnoses (κ_w = 1.00 and κ_w = 0.62) and at the level of scales (r = 0.84 and r = 0.79) as good. The internal consistency of the algorithm scale for modules 1–4, with values from r = 0.78 to 0.89, was also acceptable to good. The validity/diagnostic convergence with the Autism Diagnostic Interview-Revised (ADI-R) was 79% (κ = 0.23).

Autism Diagnostic Interview - Revised

The gold standard of ASC diagnostic procedure combines ADOS-2 with the ADI-R (Bölte et al., 2006). The ADI-R consists of a semi-structured caregiver interview. Ninety-three items investigate current ASC-typical behaviors and developmental history. The interview took place in absence of the child and was applied for all 45 participants with suspected ASC. The ADI-R diagnostic algorithm consists of the following subscales: (1) Qualitative Abnormalities in Reciprocal Social Interaction (QARSI), (2) Qualitative Abnormalities in Communication, (3) Restricted, Repetitive, and Stereotyped Patterns of Behavior, and (4) Abnormality of Development evident at or before 36 months.

Concerning the psychometric properties of the ADI-R, Bölte et al. (2006) report for the German version the following: regarding the interrater reliability for 27 of 36 algorithm-related items kappa values were κ > 0.70 (for the English original between r = 0.63 and r = 0.89 for the items of the diagnostic algorithm and r > 0.92 with regard to the scale scores of domains A–C). Retest reliabilities for the English version were between r = 0.93 and r = 0.97 for the scale scores of the domains.

Social Responsiveness Scale

The "Social Responsiveness Scale" (SRS, Bölte and Poustka, 2008) is a questionnaire of 65 items on social, communicative and rigid behavior in children and adolescents on a 4-point rating scale (1 = not true; 2 = sometimes true; 3 = often true; and 4 = almost always true). It is used for dimensional diagnostic and severity assessment of autism spectrum disorders or clarification of comorbid autistic traits in other clinical groups. It is completed by a caregiver of the respective child. Item 16, for example, addresses eye contact ("Avoids eye contact or has unusual eye contact.").

Concerning the psychometric properties of the German version of the SRS, Bölte and Poustka (2008) report the following:

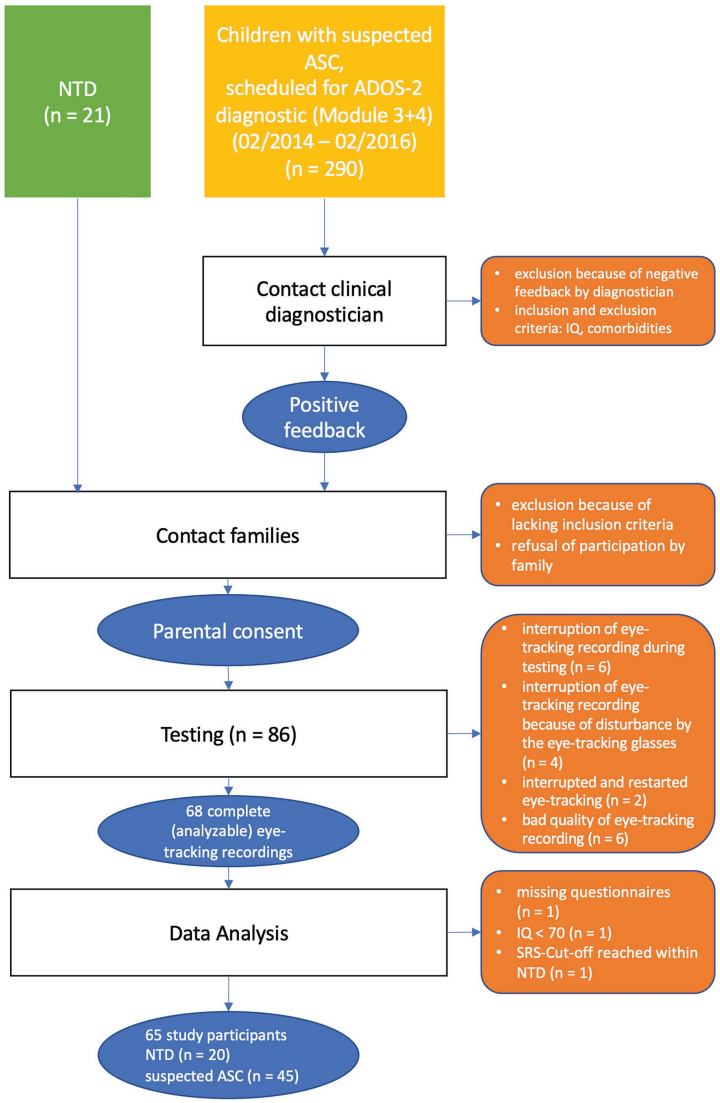

FIGURE 1 | Flow of participants.

retest reliabilities [norm sample with a time interval of 3 weeks–4 months: $r = 0.80$ for mother SRS ($N = 107$); $r = 0.72$ for father SRS ($N = 76$); mixed clinical sample for a time interval of 3–6 months: $r = 0.95$ ($N = 49$)] and internal consistencies ($\alpha = 0.93$ for mother SRS, $\alpha = 0.91$ for father SRS, and $\alpha = 0.97$ for the mixed clinical sample). The convergent validity (examined on subsamples of the mixed clinical sample) with established instruments is mediocre: ADI-R ($N = 113$): subscale social interaction: $r = 0.46$; subscale communication: $r = 0.40$; subscale stereotypical behavior: $r = 0.38$; ADOS scale communication and social interaction ($N = 119$): $r = 0.35$.

IQ Assessment

As part of the diagnostic procedure for autism spectrum disorders, nearly all participants took an intelligence test. For two participants in the ASC group, externally assessed IQ scores were not available. Because both of them were attending regular schools without difficulties, we kept them in the study.

For the additional control group, the CBCL/4–18 (to exclude psychiatric comorbidity) and an intelligence test (CFT 20-R; in order to match with the clinical groups) were completed.

Facial Emotion Monitoring

The Facial Emotion Monitoring (FEMO) is an instrument developed in-house with the goal of rapidly surveying emotional behavior (facial expression and gesture) of participants by an independent rater during the ADOS diagnostic process. Relevant aspects are inquiries about social interaction and its quality, emotional expression, and psychomotor activity. The rating takes place within the standard situations specified by the items of the ADOS-2. The FEMO assessment sheet was compiled by an independent observer based on a video recording of

the ADOS-2 to assess and rate facial and gestural expression, quality of social interaction, and psychomotility. Item 8a, for example, asks the observer to rate the assertion "The subject shows eye contact during the observation unit" on a 4-point rating scale.

Procedure: Eye-Tracking During the ADOS-2 Session

The investigation with eye-tracking glasses took place in the framework of a regular ASC outpatient diagnostic procedure, using the gold standard diagnostic instruments apart from ADI-R (Bölte et al., 2006) and IQ assessment. Out of the clinic's regular team for diagnostics of Autism Spectrum Disorder, 24 different ADOS interviewers (four male and 20 female) conducted the ADOS-2 in the study.

The Autism Diagnostic Observation Scale (Hus and Lord, 2014) serves as a basis for the acquisition of eye-tracking data. As the examined participants were exclusively children and adolescents from 8.0 to less than 18.0 years of age, with an IQ above 70, and with command of language as well as language fluency, only modules 3 and 4 were applied. For the acquisition of eye-gaze-behavior, we used the integrated interview activity *Conversation and Reporting* that is part of both modules. Following a short break, eye-tracking data were recorded during the second part of the ADOS-2 procedure. The participant put on the eye-tracking glasses; the examiner checked the correct position of the glasses and completed a three-point calibration ensuring valid recording of data before the interview began. Since accuracy is better if the calibration targets and the relevant stimuli are within the area encompassed by the calibration points (Holmqvist and Nyström, 2011), we defined the calibration points as a triangle around the visible region of the examiners body (head and upper part of the body).

In order to provide a framework for analysis, the first question in the interview section was defined as the beginning, while the participant's last answer to the last question of the interview section was defined as the end of the sequence. Lengths of analyzed video segments varied between 5 min 38 s and 44 min 0 s (NTD: M = 923 s, SD = 241 s; non-ASC: M = 1,345 s, SD = 561 s; ASC: M = 1,345 s, SD = 561 s).

Eye Movement Laboratory Procedures

Visual fixation patterns and dwell times were measured with eye-tracking equipment using hardware and software engineered by SMI (Teltow, Germany). The eye-tracking technology is video-based and uses dark-pupil/corneal reflection technique with eye-movement data collected at 60 Hz with binocular eye-tracking and integrated audio. The spatial resolution is 0.1°, and the gaze position accuracy is 0.5°. The eye-tracking glasses resemble ski-glasses, including an HD-Camera in the nose-bridge and binocular infrared sensors on the inside of the eye-glass frame. Thus, the HD-Camera records the visual field of the participant, while the binocular infrared-sensors gather data of the eye movements.

Pre-processing of Gaze Data

Definition of Areas-of-Interest

In this study, we use percentages of total dwell time as the key measure to test our hypothesis. Total dwell time describes the cumulatively calculated duration of all fixations in relation to an AOI.

For the empirical investigation of our hypothesis, we defined the following AOI: the eye region, including the left and the right eye of the interviewer not including the nasal root between them (EYES); the nose (NOSE); the mouth region of the interviewer (MOUTH); a circular area of interest in the middle of the face and a circle radius of 24 mm (referring to the face of a template, see section Fixation-based Semantic Gaze Mapping), comprising parts of the eye region and the nose (CENTER-FACE). Additional AOIs were the forehead (FOREHEAD), the chin (CHIN), and the entire face (FACE). Outside the face, we defined the following AOIs: the body of the interviewer without the face (BODY w/o HEAD) and the full body including the face (BODY WITH HEAD). The surrounding space outside the body of the interviewer is defined as white space (WHITESPACE). For an illustrative example of the template and its AOIs see **Figure 2**.

Fixation-Based Semantic Gaze Mapping

Pre-processing of raw eye-tracking data was performed with the BeGaze (version 3.7) analysis program by SMI (Teltow, Germany). The defined sequences of the interview were analyzed in a precise procedure. After a preliminary screening of the whole sequence to ensure data validity and exclude technical errors, the analysis of fixations and dwell times took place. Using a template with the defined AOIs, every fixation of the participant as recorded in the interview session was transferred manually to the corresponding region of the template face (a procedure called as "semantic gaze mapping"). The selected template is a representative front-shot of one of the diagnosticians who conducted the ADOS-2. Evaluation of the data was performed by blinded raters who had no information about group membership or diagnosis and had no knowledge about the coordinates or topography of the defined AOIs.

Statistical Analysis

Because many AOI-related dwell times (and derived measures) are not stochastically independent from each other (some AOIs overlap or are even proper part of the other), no overall ANOVA with repeated measurements with AOI as dependent factor could be computed. To put special emphasis on differences between each clinical group (ASC or non-ASC) and the NTD control group, simple one-way ANOVAs between each clinical sample and the NTD group were conducted for each AOI. Effect sizes are reported in terms of standardized mean differences (SMD). Hedges's g, rather than Cohen's d, is used as an unbiased point estimator of effect sizes (Borenstein et al., 2009), because the former enables the computation of the 95% CI. These values are also the basis of the forest plot that provides a comprehensive review of the results.

FIGURE 2 | Illustrative example of the main AOIs: 1. MOUTH, 2. NOSE, 3. CENTER-FACE, 4. FACE, 5. FOREHEAD, and 6. EYES

Correlational analyses were conducted as follows: between AOI-based percentages of total dwell times with SRS scales, Pearson correlations were computed. Correlations with items concerning quality/frequency of eye contact in the SRS, ADOS-2, and FEMO instruments were performed by nonparametric Spearman rank-order correlations because of different scale properties of the items (ADOS-2 B1, for example, is dichotomous, whereas item 16 of the SRS is evaluated on a 4-point rating scale).

All statistical analyses are performed with SAS software, Version 9.4 (SAS Institute Inc., Cary, NC, USA). For hypothesis testing, a significance level of $\alpha = 0.05$ was adopted.

RESULTS

Sample Characteristics

Tables 1 and **2** summarize the characteristics for all three subsamples concerning quantitative and qualitative variables.

There are no significant differences between the three groups with regard to chronological age. The same is true for IQ, although a trend can be seen [$F(2, 60) = 3.04$, $p = 0.055$] that is mainly caused by the lower mean in the non-ASC group ($M = 99.46$, $SD = 17.47$) as compared to the ASC and the NTD group ($M = 107.29$, $SD = 15.69$; $M = 109.80$, $SD = 9.48$, respectively). Regarding autistic symptomatology as assessed by the SRS, all six scales show significant differences between means (all Fs > 57, all ps < 0.0001; see **Table 1** for details). Gabriel's *post-hoc* comparisons revealed that – in all cases – the means for the NTD group differed significantly from the means of the two clinical groups. Conversely, the means of the two clinical groups ASC and non-ASC did not differ significantly. Also, for the ADI-R, no significant differences between the two clinical groups could be noted; only a trend could be seen for the domain/scale QARSI, where the ASC group had more pronounced scores ($M = 15.79$, $SD = 4.35$ vs. $M = 12.19$, $SD = 7.70$ for non-ASC; $F(1, 43) = 3.35$, $p = 0.074$). Additionally, it can be noted that our ASC group seems to show less autistic symptomatology, because the scores were all lower than the one reported in the ADI-R manual by Rutter et al. (2003, Table 4, pp. 44–45): The corresponding values of their validation study are QARSI: $M = 19.00$, $SD = 3.76$; QAC: $M = 16.33$, $SD = 2.96$; RRSPB: $M = 4.92$, $SD = 1.80$.

Concerning the ADOS-2, it is not possible to report scale scores, since Module 3 and Module 4 have different items, different subscales, and different algorithms resulting in incommensurable scores. Therefore, we can just report the frequencies of the three ADOS-2 diagnoses "autism" (cutoffs: M3: 9; M4: 10), "autism spectrum" (cut-offs: M3: 7; M4: 7), and "non-spectrum" for the three groups: ASC: n("autism") = 9 (47.4%), n("autism spectrum") = 6 (31.6%), n("non-spectrum") = 4 (21.1%); non-ASC: n("autism") = 2 (7.7%), n("autism spectrum") = 6 (23.1%), n("non-spectrum") = 18 (69.2%); NTD: n("autism") = 0 (0.0%), n("autism spectrum") = 0 (0.0%), and n("non-spectrum") = 20 (100.0%). The frequencies of the three ADOS-2 diagnoses is significantly different between the three groups [$\chi^2(4) = 30.41$, $p < 0.0001$].

As can be seen in **Table 2**, the main ICD-10 diagnoses for the ASC group are childhood autism (F84.0: $n = 4$), atypical autism (F84.1: $n = 3$), and Asperger syndrome (F84.5: $n = 12$).

For the non-ASC group, various main diagnoses were obtained. The majority were diagnosed with hyperkinetic disorders (F90: $n = 15$), whereas other diagnoses were sparsely distributed. For six participants in the non-ASC group, the differential diagnosis of an autism spectrum disorder remained, but the diagnostic criteria had not been met at the time of the testing.

TABLE 1 | Sample characteristics for quantitative variables of chronological age, IQ, and autistic symptomatology.

	NTD (*n* = 20)		Non-ASC (*n* = 26)		ASC (*n* = 19)			
	M	SD	*M*	SD	*M*	SD	*F*	*p*
Age	12.41	2.20	12.21	2.96	11.25	2.52	1.10	0.339
IQ[1]	109.80	9.48	99.46	17.47	107.29	15.69	3.04	0.055
SRS-T-Total	36.20	8.19	78.65	10.05	81.58	10.11	145.73	<0.0001
SRS-T-Awr	42.90	8.61	73.42	10.80	72.63	11.74	57.50	<0.0001
SRS-T-Cog	41.15	6.05	75.65	11.72	75.42	12.04	75.38	<0.0001
SRS-T-Com	40.00	6.88	81.35	12.74	87.16	12.51	107.42	<0.0001
SRS-T-Mot	41.05	7.36	73.00	10.39	80.00	11.26	89.81	<0.0001
SRS-T-RRB	47.45	4.76	74.96	7.82	76.37	9.85	91.71	<0.0001
ADI-R								
QARSI			12.19	7.70	15.79	4.35	3.35	0.074
QAC			9.73	6.06	11.21	4.04	<1	
RRSPB			3.54	2.58	4.58	2.43	1.87	0.178
AbnDev			1.65	1.50	1.21	1.36	1.04	0.313

NTD, neurotypical development; ASC, autism spectrum condition; SRS, Social Responsiveness Scale; Awr, social awareness; Cog, social cognition; Com, social communication; Mot, social motivation; RRB, restricted interests and repetitive behavior; ADI-R, Autism Diagnostic Interview-Revised; QARSI, Qualitative Abnormalities in Reciprocal Social Interaction; QAC, Qualitative Abnormalities in Communication; RRSPB, Restricted, Repetitive, and Stereotyped Patterns of Behavior; AbnDev, Abnormality of Development evident at or before 36 months.

[1]Two missing IQ values for two boys in the ASC group.

TABLE 2 | Sample characteristics for the qualitative variables gender, main diagnoses, and co-morbid diagnoses.

	NTD (*n* = 20)		Non-ASC (*n* = 26)		ASC (*n* = 19)	
	n	%	*n*	%	*n*	%
Gender (f:m)	3:17	15.0:85.0	3:23	11.5:88.5	2:17	10.5:89.5
Main diagnosis	None		F90.[0;1]: 15		F84.0: 4	
			F43.2: 2		F84.1: 3	
			F32.2: 1		F84.5: 12	
			F81.2: 1			
			F92.0: 1			
			F92.8: 1			
			F93.2: 1			
			F94.0: 1			
			F98.8: 1			
			No Fxx-diag: 2			
Co-morbid diagnoses	None		Symptoms of AD(H)D or F90.0/F90.1: 1		Symptoms of AD(H)D or F90.0/F90.1: 8	
			F98.0: 3		F43.2: 2	
			F98.8: 3		F32.1: 1	
			F43.2: 2		F81.0: 1	
			F80.0: 2		F81.3: 1	
			F95.2: 1		F82: 1	
					F95.2: 1	
					Q86.0: 1	

F32.1, moderate depressive episode; F32.2, severe depressive episode without psychotic symptoms; F43.2, adjustment disorders; F80.0, specific speech articulation disorder; F81.0, specific reading disorder; F81.2, specific disorder of arithmetical skills; F81.3, mixed disorder of scholastic skills; F82, specific developmental disorder of motor function; F90.0, disturbance of activity and attention; F90.1, hyperkinetic conduct disorder; F92.0, depressive conduct disorder; F92.8, other mixed disorders of conduct and emotions; F93.2, social anxiety disorder of childhood; F94.0, elective mutism; F95.2, combined vocal and multiple motor tic disorder [de la Tourette]; F98.0, nonorganic enuresis; F98.8, other specified behavioral and emotional disorders with onset usually occurring in childhood and adolescence; Q86.0, fetal alcohol syndrome (dysmorphic).

AOI-Based Results

In **Figure 3**, a forest plot of total dwell time percentages for different AOI is presented.

As can be seen in **Figure 3**, almost all descriptive statistics show that the ASC group differs more from the NTD group than the non-ASC group. For the AOIs CENTER-FACE and

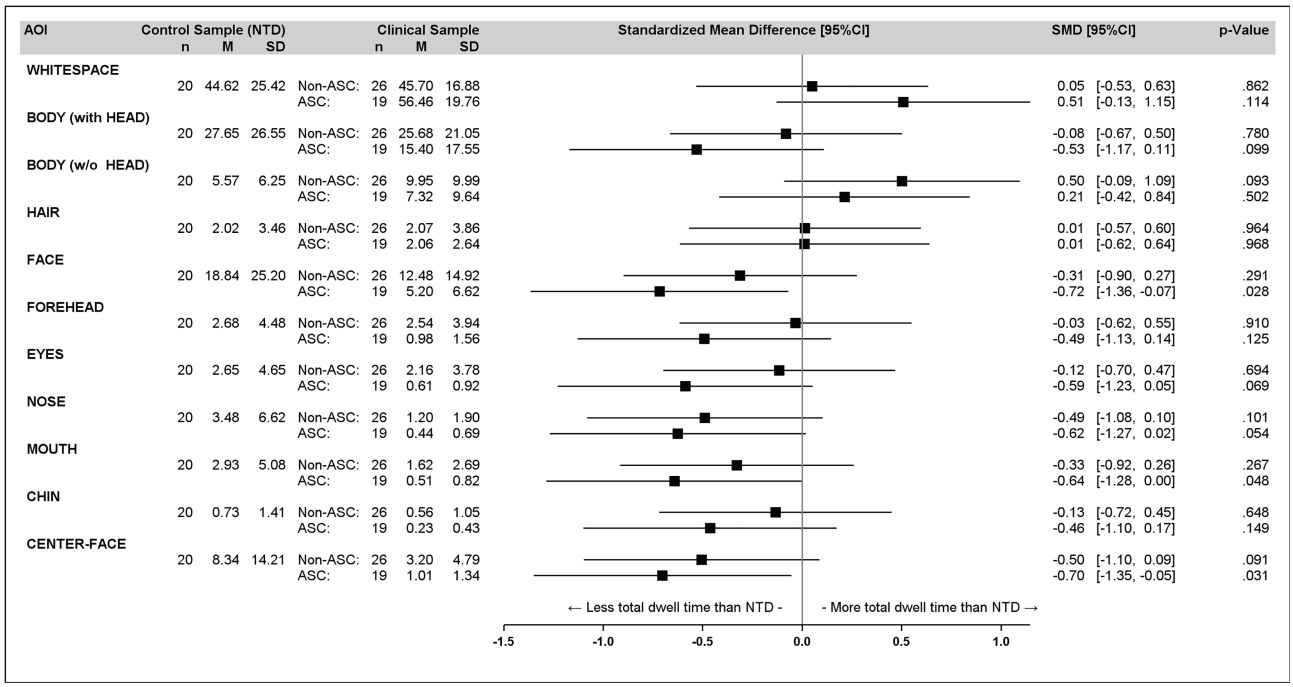

FIGURE 3 | Forest plot of total dwell time percentages for different areas of interest (AOI).

FACE, the NTDs show a significantly greater preference for the CENTER-FACE and the FACE than the ASCs [$F(1, 37) = 5.00$, $p = 0.031$, $g = -0.70$ and $F(1, 37) = 5.22$, $p = 0.028$, $g = -0.72$, respectively].

Correlational Analyses

Correlations Between AOI-Based Measures and Degree of Autistic Symptomatology

Pearson correlations between AOI-based percentages of total dwell times with SRS scales are presented in **Table 3**. There were some significant correlations (BODY with HEAD – SRS-T-Awr, FACE – SRS-T-Mot, MOUTH – SRS-T-Mot, and MOUTH – SRS-T-Total), but the most and the highest correlations were obtained for the Nose and Central Face region.

Correlations Between AOI-Based Measures and "Eye Contact"-Items From SRS, ADOS-2, and FEMO

In this section, the correlations of items concerning quality/frequency of eye contact in the SRS, ADOS-2, and FEMO instruments are presented. As a result of the different scale properties of the items (ADOS-2 B1, for example, is dichotomous, whereas item 16 of the SRS is evaluated on a 4-point rating scale), nonparametric Spearman rank-order correlations between percentages of total dwell times for the AOIs and these items were computed. All items are (re-)scaled in such a way that low values denote typical eye contact behavior, whereas higher values denote atypical eye contact. In summary, the AOI CenterFace belongs to the group with highest correlations with items concerning quality of eye contact (see **Table 4**).

Exploratory Results

Heat maps provide a quick and intuitive descriptive visual representation of eye-tracking data. They reveal the focus of visual attention and help to communicate important aspects of visual behavior.

In order to emphasize differences between all three groups visually, we created fixation-based heat maps for the first 2 min of the integrated interview activity "Conversation and Reporting." As shown in **Figure 4**, the NTD group dwells in the central face area for the longest period; the non-ASC group also shows a predominant heat pattern in the center face area. Participants on tAS, however, show no identifiable focus or long-lasting dwell time for any relevant area that is associated with para-linguistic facial expression. Looking now at the distribution of dwell times in terms of the AOI FACE, the average dwell time of the NTD group is more than three times higher than that of the ASC group, and the circular area around the nasal root (CENTER FACE) exhibit the longest dwell times within the face.

DISCUSSION

The focal point in this study was the comparison of eye-gaze behavior in individuals on tAS vs. controls in an ecologically valid standard diagnostic situation corresponding to what Schilbach (2014) calls "online social cognition". The results underline that the gaze behavior of individuals on tAS in an interactive interview situation with a real person differs from that of neurotypically developed controls. However, the differences do not seem to appear in the eye region, which is significantly

TABLE 3 | Intercorrelations of AOI-based measures and SRS scales.

	SRS Scale (T-scores)					
	Awr	**Cog**	**Com**	**Mot**	**RRB**	**Total**
AOI						
WHITESPACE	0.131	0.092	0.091	0.192	0.082	0.115
BODY w/o HEAD	–0.075	–0.104	–0.052	–0.182	–0.085	–0.099
BODY WITH HEAD	**0.256***	0.232	0.201	0.109	0.189	0.211
HAIR	–0.064	–0.060	–0.023	–0.103	–0.101	–0.056
FACE	–0.207	–0.230	–0.160	**–0.253***	–0.175	–0.213
FOREHEAD	–0.103	–0.100	–0.073	–0.167	–0.083	–0.086
EYES	–0.139	–0.176	–0.083	–0.132	–0.076	–0.113
NOSE	**–0.254***	**–0.254***	–0.229	**–0.268***	–0.207	**–0.259***
MOUTH	–0.227	–0.217	–0.203	**–0.276***	–0.185	**–0.252***
CHIN	–0.049	–0.033	–0.060	–0.134	–0.096	–0.089
CENTER-FACE	**–0.269***	**–0.278***	–0.240	**–0.284***	–0.220	**–0.273***

*SRS, Social Responsiveness Scale; Awr, Social Awareness; Cog, Social Cognition; Com, Social Communication; Mot, Social Motivation; RRB, Restricted Interests and Repetitive Behavior. *p < 0.05; significant correlations are shown in bold.*

TABLE 4 | Spearman rank-order correlations of AOI-based measures items concerning quality of eye contact.

	SRS-I16	**ADOS-2 B1**	**FEMO I8a**
WHITESPACE	0.027	**0.468*** **	**0.421*** **
BODY w/o HEAD	0.038	**–0.293***	**–0.343****
BODY WITH HEAD	0.178	–0.151	–0.124
HAIR	–0.048	–0.168	–0.098
FACE	–0.031	**–0.319****	**–0.398****
FOREHEAD	0.009	–0.200	–0.209
EYES	–0.086	**–0.298***	**–0.367****
NOSE	–0.073	**–0.253***	**–0.445*** **
MOUTH	–0.036	–0.244	**–0.386****
CHIN	0.125	–0.141	–0.243
CENTER-FACE	–0.100	**–0.281***	**–0.421*** **

*SRS-I16, Social Responsiveness Scale – Item 16 = avoiding eye-contact; unusual eye-contact; ADOS-2 B1, unusual eye-contact; FEMO I8a, FEMO Item 8a = eye-contact. *p < 0.05; **p < 0.01; ***p < 0.001; significant correlations are shown in bold.*

FIGURE 4 | Heat map of fixations for the three groups (red box left side: NTD; red box right side: non-ASC; big picture: ASC) for the first 2 min of the ADOS-2 "Conversation and Reporting" activity.

less frequented by individuals on tAS in offline social cognition tasks. While there are indeed descriptive differences, statistically significant differences in eye-gaze behavior were confirmed by dwell times in the face, the mouth and in the central face region.

There is a large body of literature on the impact and relevance of direct eye contact for social cognition (Jones and Klin, 2013; Senju, 2013; Hietanen, 2018). Many authors suggest that direct gaze plays a dominant role in social communication (Csibra and Gergely, 2006) and that the white sclera – a unique characteristic that distinguishes human beings from other primate species – is an evolutionary development to improve the basic forms of human communication (Kobayashi and Kohshima, 1997; Farroni et al., 2004; Johnson, 2005). On the other hand, the eyes themselves are by no means an exclusive source for precise information about identity, emotional state, or mood of the observed person, quite apart from the fact that long-lasting eye contact elevates physiological arousal (Nichols and Champness, 1971) and provokes expectations of behavior.

One could thus presume that a balanced mix of mutual social interaction evinces a structured pattern of gaze sequences, which enables the interacting partners to read information efficiently from the face of the counterpart. In the field of reading research, there is evidence for preferred landing positions (PLP; Rayner, 1979) in sentence reading and of optimal viewing positions (OVP; O'Regan et al., 1984) in isolated word recognition (for a recent review, see Hyönä and Kaakinen, 2019). Given this background, the research efforts in object recognition identified similar PLPs and OVPs for optimal recognition performance (Foulsham and Kingstone, 2013).

The significant differences between NTD and ASC in our sample particularly in the AOI CENTER FACE reveal longer dwell times in the middle of the observed face in the NTD group. This arguably indicates that neurotypically developed face readers use this region as an optimal viewing position for successful categorization of facial expression.

In comparison with current studies it can be said that there are both representatives of an unimpaired holistic face categorization in ASC (Tanaka and Sung, 2016; Ventura et al., 2018) as well as researchers who assume an impairment in

holistic face processing (Brewer et al., 2019). In an older offline study by Tanaka et al. (2012) it was found that individuals on tAS have a tendency to recognize the mouth region holistically, but the eyes as an isolated part of the face.

Apart from this many recent studies with an offline design suggest that emotion categorization is impaired in ASC (Uljarevic and Hamilton, 2012; Lozier et al., 2014; Velikonja et al., 2019). An eye-tracking study conducted in 2019 considered the question of how atypical face processing is related to differences in visual conjunctive processing (Stevenson et al., 2019). The study revealed that increasing ASC symptoms are associated with reduced levels of conjunctive processing. Although this offline study used photographs of virtual faces and the authors suggest untypical visual conjunctive processing in ASC, there are no indications for a starting point of conjunctive face processing in ASC.

Notably, Hsiao and Cottrell (2008) found that an optimal position for face recognition is around the center of the nose. However, this is contrary to the results of a large number of lab studies, which indicate that the eyes and the mouth region are highly relevant for face recognition. This suggests that there are differences in PLP and OVP between offline lab studies and real online social interaction (Foulsham, 2020). One reason for this may be that the online character of ecologically valid social situations has other prerequisites than an offline experiment with a precisely defined task. Constructive and active participation in a real-time social interaction is associated with a different approach to cognitive processing that is characterized by reciprocal relations as opposed to situations in which social phenomena are merely observed (De Jaegher, 2008; Schilbach, 2010; Wilms et al., 2010).

The reciprocity of social interactions demands an implicit repertoire of rapid and flexible processes in a circular operational sequence of action and reaction. Whereas offline social cognition is only based on an observer position without the additional cognitive load of being involved in an interaction, the participant in a socially interactive process is only able to react adequately if the constantly flowing information can be categorized in the context of the developing situation and in compliance with his own social schemas (Schaller and Rauh, 2017; Schaller, 2019). In order to make efficient use of the face of the counterpart, one must possess implicit face-detection strategies, capturing all relevant hints for a better understanding of the social situation.

Looking now at the visual scan pathways of the three groups, it can be ascertained that the NTD group in particular shows significantly longer dwell times for the circular area around the nasal root (CENTER FACE). This is astonishing, because a direct gaze in the eyes of the counterpart occurs less frequently than on the forehead or the mouth.

It follows, therefore, that with regard to the distribution of the AOIs in the face, the main focus is not in the eyes. Instead, there is evidence that the region around the nose is the most visited and revisited area of interest in the face. NTD tend to dwell eight times longer in the center face area than the ASC group. This phenomenon can be visually presented by comparing the heat maps of both groups. While the NTD group develops a clear center face preference in the heat map within a timeframe of less than 2 min (see **Figure 4**), the distribution of fixations in the ASC group shows an unstructured spread of seemingly uncoordinated scanpaths without a clear focus on any of the relevant AOI for facial information.

The majority of fixations in the ASC group lie outside the face or in parts that do not provide any information about facially expressed emotions (hair, ears, and chin). Based on this result, we suggest that neurotypically developed individuals have an implicit automatism, using the center of the face as the Archimedean Point from which the facial expression can be gathered as a valid source of information. Furthermore, our results are supported by Bobak et al. (2017), who showed that the dwell times on the eye region did not correlate with face perception skills of controls, while there was a significant and robust correlation between the ability to recognize faces and dwell time spent on the nose.

A further indication for the tendency to use the center of the face as optimal viewing position for a better recognition and categorization of facial expression can be found in so called "Super Recognisers," who outperform neurotypical individuals in face recognition (Russell et al., 2009). Individuals who meet the criteria for super recognition use the nose instead of the eyes to achieve an efficient distribution of spatial attention across the face, resulting in higher-than-average face recognition (Bobak et al., 2017).

The significant differences between groups concerning the mouth region are consistent with the findings in offline social cognition that individuals on tAS spend less time on the mouth region as compared to their neurotypically developing peers (Wagner et al., 2013).

Turning to an analysis of correlations between AOI-based measures and the degree of autistic symptomatology, the AOIs NOSE and CENTER FACE reveal the highest correlations with regard to social responsiveness, in so far that high rates in the SRS total score result in shorter dwell times for CENTER-FACE.

Looking particularly at the AOI CENTER-FACE, we find the highest correlations with the SRS subscales Social Cognition and Social Motivation. The subscale Social Cognition is defined as the ability to adequately interpret social key stimuli, while the subscale Social Motivation reflects the need for social interaction. It is, therefore, a fair assumption that the use of the center face as an ideal basis for implicit face-detection strategies is a relevant criterion for social interaction abilities.

Furthermore, the correlation between AOI based measures and the ADOS-2/B1 item "unusual eye-contact" initially shows an expected pattern of significant positive correlation concerning WHITESPACE and a significant negative correlation with respect to EYES. However, there are also significant correlations for the AOIs NOSE and CENTER-FACE. The highest correlation can be found for FACE and ADOS-2 B1, which suggests that, from the rater's perspective, a participant's glance in the face of the diagnostician can be a sufficient indicator for neurotypical eye-contact. On the other hand, it raises the question as to whether the rater is able to differentiate between actual mutual gaze and a fixation of the nasal root or one of the eyebrows.

In order to have a third-party assessment of the ADOS-2 "Conversation and Reporting" activity in terms of emotional

behaviors, such as facial and gestural expression, quality of social interaction, and psychomotility, we compiled the FEMO. Initially, it was used to prove the extent to which the ADOS rater assessment corresponds with the FEMO observation and the eye-tracking data. In this context, item 8a is particularly important, with high values denoting unusual and minimal eye-contact. Here, we also find the highest significant negative correlations in the AOIs NOSE and CENTER FACE and a highly significant positive correlation for the AOI WHITESPACE.

Thus, it can be concluded that gaze behavior in an ecologically valid online social situation clearly differs from offline situations.

Limitations

The limitations of the study are, first, that current results are based on a relatively small sample. Therefore, only large effects could be detected. In order to generate more conclusive data that can detect small and medium effect sizes, it would be appropriate to develop study concepts involving large numbers of participants.

Concerning the statistical analyses, the multiple comparisons' problem arose (1) in analyses concerning group differences for the various AOIs and (2) in the correlational analyses. Because of lack of stochastic independence for the total dwell time-related measures in (1), no justifiable adjustments for alpha could be made. For the correlational analyses, we ran the analyses without alpha adjustments, since there is no gold standard how to deal with the multiple comparisons' problem – a problem that is still under debate (e.g., Rubin, 2017), and for which Bonferroni correction seems to be a suboptimal solution having its own problems (e.g., inflation of type II errors; see Perneger, 1998). Therefore, the question of whether our results are reliable should be answered by replication studies.

Thirdly, we only tested children and adolescents from 8 to less than 18 years of age. A wider spectrum of age ranges, including younger children, adults, and older participants, could offer further information about the development of gaze behavior during online social situations in participants with and without ASC.

Furthermore, this sample had a negligible proportion of female participants, so that no gender-specific differences could be evaluated for. Future investigation of gaze behavior may help to find gender specific differences.

In this study, we chose to focus on the gaze behavior of the ASC group. The non-ASC group turned out to be a very heterogeneous cohort, with too many different diagnoses to run additional analyses.

Moreover, we only investigated individuals on tAS without intellectual disabilities, which makes it impossible to generalize the results for all individuals on tAS. With regard to autistic symptoms, our group is more likely to show less pronounced severity. Thus, the extent of untypical gaze-behavior in our ASC group may underestimate the real extent of deviating gaze-behavior in people with ASC who do not have co-occurring intellectual disabilities.

Lastly, we used eye-tracking data without any other psychophysiological parameters. Future research in online social cognition might combine eye-tracking and psychophysiological measures in order to clarify any existing correlations.

Conclusion

With the face being a projection surface for expression, its interpretation is dependent on the spectrum of a performer's facial expressions and the repertoire of emotional expressive categories and social schemas available to the observer. The central face seems to be the hot spot, where many socially relevant behavioral expressions as well as social information perceptual processes meet.

Additionally, contextual factors, like underlining gestures, body movements, paraverbal signs, and sceneries, specifically influence the perception and categorization of facial stimuli (Aviezer et al., 2017). Thus, it is important to keep track of the counterpart's face while considering contextual variables or the general setting of a certain social situation (Pfeiffer et al., 2013).

The results of this study show that it is not the eyes but the central face region that is an important anchor point in using all the above-mentioned factors efficiently.

While in neurotypical individuals this implicit and procedural development takes place in an emergent process of exchange with the social environment, it seems that this development is different in individuals on tAS.

Consequently, it will be necessary to analyze this process in further studies of online social interaction, particularly by comparing factors such as contextual background and social schemas. In parallel, a clinical study with enlarged number of participants and a broader age range, considering children, adolescents and adults is under way.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because of confidentiality reasons. Requests to access the datasets should be directed to reinhold.rauh@uniklinik-freiburg.de.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of the University of Freiburg. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

US, RR, and CF developed the study concept and designed the study with assistance of AB, AI, and MH. AB, AI, and MH coordinated the study, recruited the participants, and completed data collection. AB and AI checked the integrity and the accuracy of the gaze data and performed the gaze data pre-processing pipeline with supervision of US and RR. RR planned and carried out the statistical analyses with

contributions of US and MH. All authors made contributions to the interpretation of the data. US, RR, and AB drafted the initial manuscript with contributions of AI, CF, MB, MH, and LT. All authors reviewed and revised the manuscript and approved the submission of the final manuscript.

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Autistic Self-Advocacy and the Neurodiversity Movement: Implications for Autism Early Intervention Research and Practice

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The growth of autistic self-advocacy and the neurodiversity movement has brought about new ethical, theoretical and ideological debates within autism theory, research and practice. These debates have had genuine impact within some areas of autism research but their influence is less evident within early intervention research. In this paper, we argue that all autism intervention stakeholders need to understand and actively engage with the views of autistic people and with neurodiversity as a concept and movement. In so doing, intervention researchers and practitioners are required to move away from a normative agenda and pay diligence to environmental goodness-of-fit, autistic developmental trajectories, internal drivers and experiences, and autistic prioritized intervention targets. Autism intervention researchers must respond to these debates by reframing effectiveness, developing tools to measure autistic prioritized outcomes, and forming partnerships with autistic people. There is a pressing need for increased reflection and articulation around how intervention practices align with a neurodiversity framework and greater emphasis within intervention programmes on natural developmental processes, coping strategies, autonomy, and well-being.

Keywords: autism, children, neurodiversity, self-advocacy, early intervention

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INTRODUCTION

The last two decades have brought about huge socio-political shifts within the world of autism theory, research and practice. In the mid-1990s, the emergence of the internet provided a more accessible text-based means of communication and empowered a growing number of autistic people to connect and share ideas with one another (Dekker, 2020)[1]. Out of the early autistic social groups of the 1990s emerged autistic culture, the autistic self-advocacy movement, and the assertion that autism is a valid way of being. This environment also gave rise to the neurodiversity movement (Singer, 1998). Through the 2000s, the neurodiversity movement has been galvanized in a large part due to the voices, advocacy and protest of the autistic community, facilitated through

[1]We use identity-first language in keeping with the preference of many autistic people (Kenny et al., 2016). We also use the more neutral term "autism" rather than "Autism Spectrum Disorder".

developments in online communication and networks (Kras, 2009) and is increasingly influencing academic, clinical and lay understanding of autism and other forms of neurological difference.

A central premise of the neurodiversity movement is that variations in neurological development and functioning across humans are a natural and valuable part of human variation and therefore not necessarily pathological (e.g., Jaarsma and Welin, 2012; Kapp, 2020). Neurodiversity as a social justice and civil rights movement intersects with the wider disability rights movement (Hughes, 2016). The most significant premise of both is that disability is not simply a defect in the individual, but arises from the interaction between a non-standard individual and an unaccommodating environment (the social model of disability; Oliver, 1990). Consistent with this stance, many neurodiversity proponents do view autism as a disability. From this theoretical underpinning, the neurodiversity movement makes several demands, including the recognition and acceptance of the value of cognitive variation as a form of biodiversity and hence its positive contribution to groups, communities and societies (the social-ecological perspective; Chapman, 2020) and equal rights leading to an end to discriminatory policies and practices (Runswick-Cole, 2014).

The amplification of autistic viewpoints, coupled with the traction of neurodiversity as a concept and movement, has led to the emergence of new ethical, theoretical and ideological debates. These debates and discussions have had genuine impact within some areas of autism research, predominantly that focused on adults. Examples of this impact include: (a) debates over whether the social difficulties experienced by autistic people are best understood as being a problem within the individual, or a problem between two (mis-matched) individuals, and the resulting research into the Double Empathy Problem and diversity in social intelligence (Milton, 2012; Crompton et al., 2020); (b) calls from the autistic community for a greater emphasis on improving mental health and quality of life in autistic individuals (Autistica, 2015; National Autistic Taskforce, 2019) and an increase in research into effective, person-centered mental health interventions (e.g., Crane et al., 2019; Cassidy et al., 2020; Parr et al., 2020) and (c) research into community preferences over the language used to describe autism and autistic people (e.g., Kenny et al., 2016; Bury et al., 2020). Despite these impacts within adult-focussed research, these debates are still rarely directly addressed in early intervention research, where the impact of the autistic viewpoint is often implicit or not present at all. The absence of clear and proactive engagement with these debates contributes to a lack of confidence in an evidence base that has already shaky foundations due to its poor methodological quality and widespread unreported conflicts of interest (e.g., French and Kennedy, 2018; Bottema-Beutel et al., 2020; Sandbank et al., 2020). In this paper, we argue that all autism intervention stakeholders need to understand and actively engage with these debates. We focus on psychosocial intervention programmes that aim to improve aspects of young autistic children's cognitive, behavioral, emotional, or relational functioning and reflect on the purpose of early autism intervention, the types of intervention methods we use, and how these align with the priorities of autistic people. We then reflect upon issues pertinent to research into early autism intervention and the challenges and opportunities presented by these shifts, pointing to future directions.

IMPLICATIONS FOR EARLY AUTISM INTERVENTION

Whilst there is agreement amongst most of the autistic community, clinicians, and researchers that interventions should be available to help autistic people of all ages to thrive and reach their potential (UK Parliament, 2020), there are many controversies surrounding what this means in practice. Until the 1990s, it was common to consider therapy for autistic children as a means of reaching the child within their "autistic shell" and drawing them out, resulting in a normal or near-normal child (e.g., Park, 1972; Kaufman, 1976; Maurice, 1998). The earliest actions of the autistic self-advocacy movement were to call for the recognition of autism as an essential aspect of the person (Sinclair, 1993). Autistic self-advocates opposed early autism interventions with a stated treatment goal to make a child no longer, or less, autistic. However, some stakeholders, in particular parents of autistic children with substantial intellectual, language and behavioral challenges, argued that autistic adults without these challenges could not speak to their children's experience, and that their children required such interventions in order to achieve a reasonable quality of life (Dekker, 2017; Fletcher-Watson, 2018). This disagreement has yet to be fully resolved. Some activists continue to argue that any attempt to alter an autistic person is misguided, thereby rejecting any form of early intervention (e.g., Stevenson, 2015). Some autistic people, parents or other stakeholders continue to oppose neurodiversity as a concept or social movement, arguing, for example, that it presents a sanitized view of autism, excludes those with significant language or intellectual disability, and deflects resources from those most in need of support (Happé and Frith, 2020; Hughes, 2020).

Objections to neurodiversity are often based on an erroneous conception of the tenets of the movement (den Houting, 2019). Fundamentally, neurodiversity emphasizes the collective strength inherent in cognitive diversity (Chapman, 2020) and that this strength arises from all kinds of differences, including those associated with autism, intellectual disability or language impairment (Kapp, 2020). Moreover, neurodiversity activism, which includes some non-speaking activists, specifically includes and advocates for those who are unable to do so themselves. A balanced view of neurodiversity recognizes that, whilst diversity brings fundamental collective advantages, within any one neurodivergent individual weaknesses are often the inextricable partner of strengths, and that individuals can want things to be different and still want to be themselves. It includes the understanding that some neurological differences are disadvantageous, either inherently or in interaction with the environment, and could benefit from correspondingly targeted intervention.

Adopting this balanced account of neurodiversity, we can derive three important implications for intervention. Firstly, neurodiversity-informed intervention opposes any attempt to "cure" or "normalize" autistic children, and, whilst in many contexts this talk is no longer acceptable (Happé and Frith, 2020), there are still many interventions purporting an explicit or implicit curative or normative agenda (Mottron, 2017). This opposition is conceptual: even if it were desirable, it would not be possible to cure someone of an innate neurological difference. It is also existential: autism is so pervasive and profound, that attempts to target autism itself fundamentally changes the person; many autistic people have equated being cured of autism as tantamount to death, as they would be a completely new individual (Sinclair, 1993). There is also increasing evidence to support opposition on ethical grounds as: (a) this approach leads to individuals "masking" their autism or attempting to "pass" as neurotypical at a huge cost to their mental health and well-being (Milton and Moon, 2012; Mandy, 2019) and (b) many intervention programs attempt to teach "normative behavior" without referencing empirical evidence for what "normative behavior" looks like and thereby teach autistic children to behave in ways that do not actually resemble autistic or non-autistic children (Bottema-Beutel et al., 2018).

Secondly, interventions informed by neurodiversity do carefully address any extrinsic factors around an autistic child that contribute to disadvantage and negative experiences and therefore aim to improve the "goodness of fit" between the child and their physical or socio-emotional environment (Lai and Szatmari, 2019). Interventions that encourage and provide opportunities for physical, sensory and emotional regulation (e.g., sensory integration therapy, Randell et al., 2019) are compatible with this stance. Interventions can promote an understanding of autism and neurodiversity in people in the child's world, such as caregivers (e.g., EMPOWER-ASD intervention,[2] Systemic Autism-related Family Enabling intervention, McKenzie et al., 2019; SOLACE programme, Lodder et al., 2020), and education professionals and peers (e.g., Learning about Neurodiversity at School project[3]). These interventions also support non-autistic people to build resilience, develop a positive philosophy toward the autistic child, and to build relationships in a respectful, supportive and harmonious manner. Targeted interventions for autistic children can also build effective communication between the child and others, for example, by coaching caregivers and education professionals to "speak the child's language" (e.g., Paediatric Autism Communication Therapy; Pickles et al., 2016; Green et al., 2018). Other interventions aim to support neurodivergent children by working with them directly to understand their autism and build self-awareness and self-esteem (e.g., Pegasus, Gordon et al., 2015; the Spectacular Girls programme[4]). Intervention efforts that target the child's environment may address early external causes of distress (e.g., non-acceptance/non-accommodation of needs, bullying, and exclusion) and therefore help to prevent future mental health problems.

A third implication for interventions concerns those aspects of autism that are disadvantageous in and of themselves. A balanced view of neurodiversity mandates that specific characteristics of autism be depathologised, unless those characteristics cause harm or discomfort to the individual or a violation of others' rights. The complexity for autism interventions concerns the fine line between supporting a child's development and attempting to change the essence of the person. It also concerns the fine balance between accommodation of autistic behaviors and the alleviation of actually or potentially detrimental cognitive or behavioral phenomena. This balance is challenged by differing opinions as to what constitutes and causes suffering, and difficulties in ascertaining the views of individual children due to their young age, communication difficulties, and lack of understanding of potential future consequences. There are no simple solutions to these complexities. However, there are some principles that can guide us in a direction that is consistent with autistic viewpoints and a neurodiversity stance.

Consideration of Internal Drives and Experiences

A key principle concerns looking beyond observable behavior to consider internal drives and experiences. An under-appreciation of the sensory and emotional experiences of neurodivergent children can result in attempts to reduce or eliminate natural coping and self-regulation strategies, such as repetitive motor mannerisms or "stimming" behaviors (Bascom, 2012; Kapp et al., 2019). Eliminating such behaviors can lead to children being unable to avoid aversive experiences, calm themselves, or to communicate intense emotions (Kapp et al., 2019). Moreover, there is increasing evidence that different developmental routes can lead to the same outcome, whereby atypical developmental processes are actually beneficial to that individual's intrinsic developmental trajectory; examples are echolalia and hyperlexia as alternative routes into functional spoken language (Mottron, 2017). Focussing on reduction of the behaviors that define the autism diagnosis fails to consider that these behaviors are the outcome of different underlying neurology and interfering with them may undermine a child's natural coping strategies and development. Early interventions should therefore work with (not against) the child's developmental trajectory, as well as with their natural way of learning (Fletcher-Watson, 2018).

Re-evaluation of Intervention Targets

We should evaluate the motivations driving the decisions around intervention targets and not assume that the things that make a good neurotypical life are identical to autistic priorities (Buckle, 2013; Milton, 2014; Iemmi et al., 2017). Active listening to the autistic community helps understand autistic priorities around intervention targets, as does close attention to research that highlights the phenomena that cause autistic people difficulty or distress, affect quality of life and for which autistic people actively ask for support. Examples include autistic inertia (Buckle et al., 2020), life skills (Pellicano et al., 2014), intolerance of uncertainty (Rodgers et al., 2018), and anxiety (Robertson

[2]www.reach-asd.org.
[3]https://dart.ed.ac.uk/research/leans/.
[4]https://helenclarkeautism.com/spectacular-girls.

et al., 2018). Certainly, avoiding intervention techniques that themselves cause emotional harm is crucial, and a key underlying principle is to support the autistic child's ability to exert choice and control in their life as they develop.

Emphasis on Strengths, Pleasure, and Well-Being

Interventions should respect and enhance those things that bring happiness and joy. Passionate interests can bring pleasure and relaxation through repetition or intensity of immersion in tasks, behaviors or objects (e.g., autistic reflections on flow states; Murray et al., 2005; McDonnell and Milton, 2014). Predictable access to preferred activities not only decreases expressions of negative emotions (sometimes manifest as "challenging behavior") but also can provide opportunities for expertise and genuine social bonding (Mottron, 2017; Grove et al., 2018; Wood, 2019). The adoption of a positive psychology and strengths-based stance (Burnham Riosa et al., 2017; Dykshoorn and Cormier, 2019) refocuses intervention efforts away from reducing deficits and toward enhancing those activities or skills that naturally lead to learning, social connection, and well-being. Intervention efforts should leave alone unconventional characteristics that cause no harm to self or other, such as a monotone voice or preference for solitude. Lifespan research into the childhood factors that are associated with long-term well-being will enable us to boost these important factors through early intervention (Rodogno et al., 2016; Pickles et al., 2020).

Promotion of Autonomy

The final fundamental principle concerns autonomy and the right to say "no". Poignant accounts from autistic adults describe the use within early interventions of overbearing physical prompting, ignoring of communication attempts, or outright removal of their right to communicate "no" and how this left them passive, traumatized, and vulnerable to abuse (Kirkham, 2017; McGill and Robinson, 2020). These practices must be avoided. Autonomy is essential to creating the life one wants to lead (National Autistic Taskforce, 2019; Späth and Jongsma, 2020). In order to achieve any significant level of autonomy, one must have functional communication, so interventions supporting communication (not simply speech) and understanding required for the expression of autonomy are justified, as long as they are undertaken ethically, with true respect for the individual.

APPLICATION OF THE NEURODIVERSITY FRAMEWORK TO AUTISM INTERVENTION RESEARCH

Re-Framing Effectiveness

Early intervention researchers understand the importance of an evidence base and effectiveness is often the key factor when evaluating evidence. Clearly, it is critical that research informs us about intervention effectiveness – no one wants to spend limited resources on interventions that do not work. However, effectiveness needs to be understood within the context of the above principles. While an intervention may be effective at reducing autistic behavior, if it leaves the child without coping mechanisms or at risk of mental health difficulties, it has not been effective in improving their life. We need to reframe effectiveness to concentrate on the outcomes that are most important to the long-term well-being and autonomy of the children involved and the preferences and priorities of autistic people (Neumeier and Brown, 2020); research can then evaluate the extent to which these prioritized outcomes are (or are not) improved by any particular intervention.

Outcome Measurement

The landscape of tools used to measure intervention outcomes is strongly focused on the reduction of autism symptoms (e.g., Provenzani et al., 2020). Conceptually, this falls squarely within a normalization agenda: if children's autistic behaviors are reduced sufficiently, they will no longer meet the criteria for autism. In practice, autism symptomatology as a metric amalgamates many different variables. Many of these target variables are incompatible with a balanced view of neurodiversity, such as imposing non-autistic social behaviors or reducing sensory behaviors or motor mannerisms that act as coping strategies. However, others are consistent with it, e.g., improving communication (Kapp, 2020). As a discipline, we need to move measurement away from autism symptomatology and produce validated tools that assess the goodness of fit between an individual and their social, emotional and physical environment. There are good examples already, such as the Autism Five Minute Speech Sample that measures the emotional climate around the autistic child (Benson et al., 2011), and the Dyadic Communication Measure for Autism that assesses caregiver communicative synchrony (Aldred et al., 2004; Green et al., 2010). However, additional measures of environmental outcomes are needed. We also need robust and creative ways to measure, in children with all levels of communication ability, specific and transparent intervention outcomes that are verifiably beneficial, including autonomy, quality of life and the variables that easily impact on these, such as functional communication, inertia, and anxiety (McConachie et al., 2015). The International Classification of Functioning, Disability and Health's "Core Set for Autism Spectrum Disorder" (Bölte et al., 2014) assesses such outcomes within clinical contexts and could be developed for use within intervention research.

Partnerships With Autistic People

In the UK there is now an increased understanding amongst researchers and funding bodies of community priorities (e.g., James Lind Alliance, 2016) and more meaningful involvement of autistic people in research (Pellicano et al., 2014; Fletcher-Watson et al., 2019). More neurodivergent/autistic people are leading academic discourse (e.g., Chapman, 2020; Kapp, 2020) and empirical studies (e.g., Belcher et al., 2019; Buckle et al., 2020) and there is a greater emphasis on participatory and action research models with autistic viewpoints and experiences at the center (e.g., Crane et al., 2019; Lam et al., 2020). These developments have cast light on the need for autism researchers to re-align their priorities and rethink some of the ways in

which they work, thereby slowly changing the emphasis and tone of research.

Parents have historically been the default channel for meaningful involvement within research, and trials of early interventions still typically center on parental views and priorities (e.g., Leadbitter et al., 2018). Although they have their child's best interests at heart, neurotypical parents may be missing essential aspects of understanding from their autistic child's perspective. One argument put forward against involving autistic adults in child-focused research is that articulate and intelligent autistic adults cannot speak for the experience of children with significant intellectual or language disability. We need to be much more invested and creative in exploring ways to garner and document the views of children and adults who have severe communication impairments and this is an important focus for future research (Happé and Frith, 2020). We also need to recognize that autistic adults often bring valuable expertise to child-focused research. Autistic people can speak to what a good autistic life is like (Iemmi et al., 2017) and what might have helped them. Many autistic self-advocates are parents of non-speaking children or were such children themselves. Some autistic people are well-connected with others and can draw on a wide range of experiences. Researchers can also become better acquainted generally with autistic viewpoints through the sentiments actively, and often passionately, shared by autistic people in general forums. It is easier than ever before for neurotypical researchers to access and understand autistic culture and preferences through books, blogs, video accounts, and social media posts.

CONCLUSIONS AND FUTURE DIRECTIONS

Autistic self-advocacy and the neurodiversity movement offer up valuable opportunities to autism intervention practitioners and researchers. A balanced neurodiversity stance offers key principles to steer the development, delivery and evaluation of early interventions. Future directions for research and practice include: (1) partnerships with autistic people, alongside caregivers and other stakeholders, on intervention research steering and advisory boards and throughout engagement, involvement and co-production processes; (2) reflection by intervention researchers and practitioners upon how their intervention practices align with a neurodiversity framework and the views of autistic people, particularly around intervention targets and methods, and more transparent articulation of these issues in engagement and dissemination activities; (3) greater regard within intervention programmes to natural autistic developmental processes, coping strategies, autonomy and well-being; and (4) increased efforts to develop and validate tools to measure autistic prioritized outcomes and the goodness-of-fit between an autistic individual and their environment. With close attention to the needs, preferences and priorities of autistic people, we can move beyond historical divides, misunderstandings and wrongdoings to a place where we value the expertise of autistic people, embrace practices that respect and accept individual neurotypes, and ensure our interventions address the things that matter most to the recipients.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

AUTHOR CONTRIBUTIONS

All authors contributed to the development of ideas and viewpoint, the review of the literature, and writing the article.

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Local Processing Bias Impacts Implicit and Explicit Memory in Autism

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Autism spectrum disorder (ASD) is characterized by atypical perception, including processing that is biased toward local details rather than global configurations. This bias may impact on memory. The present study examined the effect of this perception on both implicit (Experiment 1) and explicit (Experiment 2) memory in conditions that promote either local or global processing. The first experiment consisted of an object identification priming task using two distinct encoding conditions: one favoring local processing (Local condition) and the other favoring global processing (Global condition) of drawings. The second experiment focused on episodic (explicit) memory with two different cartoon recognition tasks that favored either local (i.e., processing specific details) or a global processing (i.e., processing each cartoon as a whole). In addition, all the participants underwent a general clinical cognitive assessment aimed at documenting their cognitive profile and enabling correlational analyses with experimental memory tasks. Seventeen participants with ASD and 17 typically developing (TD) controls aged from 10 to 16 years participated to the first experiment and 13 ASD matched with 13 TD participants were included for the second experiment. Experiment 1 confirmed the preservation of priming effects in ASD but, unlike the Comparison group, the ASD group did not increase his performance as controls after a globally oriented processing. Experiment 2 revealed that local processing led to difficulties in discriminating lures from targets in a recognition task when both lures and targets shared common details. The correlation analysis revealed that these difficulties were associated with processing speed and inhibition. These preliminary results suggest that natural perceptual processes oriented toward local information in ASD may impact upon their implicit memory by preventing globally oriented processing in time-limited conditions and induce confusion between explicit memories that share common details.

Keywords: autism, episodic memory, priming, perception, attention

INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by atypical visual perceptual abilities with superior performance on many perceptual tasks that require local processing, including the block design task (Caron et al., 2006; Kuo and Eack, 2020), the embedded figures task (Shah and Frith, 1983; Jolliffe and Baron-Cohen, 1997), visual search (Plaisted et al., 1998; O'Riordan et al., 2001) or feature discrimination (O'Riordan and Plaisted, 2001). Several cognitive theories have been proposed to explain this superior visual search for local details. First, the "weak central coherence" account (WCC) developed by Frith and Happé postulates a weakness in integrating local details into a global and coherent form (Happé and Frith, 2006). These authors argue that this is a "cognitive style" rather than a core deficit and that a local processing bias may be overcome when explicit global processing is required. For example, it was found that persons with ASD showed lower levels of global processing on a divided attention task but not on a selective attention task (Plaisted et al., 1999; Van der Hallen et al., 2017), and recently, Avraam et al. (2019) found that, when the local and global levels are not competing, individuals with autism demonstrate robust global organization (grouping processes) that operates even when not directly instructed and questions the WCC theory. The second theory focuses on enhanced low and mid-level processes of perception that allow ASD people to detect and memorize the surface properties of visual and auditory patterns, and is summarized in the Enhanced Perceptual Functioning model (EPF, Mottron et al., 2006). This theory postulates that people with ASD have a natural bias to process stimuli locally confirmed by neuroanatomical and behavioral findings (Mottron et al., 2013a; Chung and Son, 2020). This model also implies that global configurations may be processed in a typical manner when experimental conditions are right, such as when global strategies are more appropriate for performing the task (Mottron et al., 2006). Theses both theories posit that individuals with ASD are biased toward local or featural information rather than global properties of a stimulus. These enhanced perceptual abilities may also result from atypical attentional processes (Plaisted et al., 1999; Keehn et al., 2013; Kaldy et al., 2013). Keehn et al. (2013) reported in Autism an impairment of the three independent attentional networks described in Posner and colleague's model of attention (Petersen and Posner, 2012): alerting, orienting and executive control networks. Petersen and Posner suggest that impairment of the orienting network and more specifically the resistance to attention disengagement may be at the origin of many behavioral features of ASD. Consequently, visual search superiority may be related to a tendency to over-focus coupled with abnormal attentional disengagement. In this context, the visual local processing bias may result from difficulties in shifting from salient details to the global shape. When such a shift is required, individuals with ASD would need longer to disengage their attention to perform as typical controls.

This atypical perceptual functioning may affect other areas of functioning that people with ASD find difficult such as social interactions or communication (Falter et al., 2012 for ASD people with speech onset delay). Another less investigated domain is memory, which may also be affected by this perceptual bias. An interesting distinction to explore the impact of the perceptual profile of ASD subjects is that between implicit and explicit memory. Implicit memory has been defined as the expression of past experiences occurring beyond the boundaries of consciousness and without any intentional recollection (Graf and Schacter, 1985). Priming is one of the most well-known implicit memory phenomena and refers to a change in the speed or accuracy with which a stimulus is processed following prior experience of the same or a related stimulus. Different kinds of priming have been identified, such as perceptual priming, which is based on the physical properties of the stimuli (Tulving and Schacter, 1990). The few studies that have explored this kind of implicit memory in ASD have shown intact priming effects (Renner et al., 2000; Toichi and Kamio, 2001; Gardiner et al., 2003). These priming tasks provide the opportunity to evaluate the effect of local precedence for the ASD group or global precedence for typically developing participants on memory under conditions that favor automatic processing uncontaminated by conscious attentional processes as well as promoting a participant's preferred perceptual processing style. Recently, Hine and Tsushima (2018) shown that not explicit but implicit memory was affected by the perception style (perceptual index calculated to the Navon task): local perception style people more greatly used implicit memory than global perception style people.

In contrast to preserved performance in implicit memory, several studies report difficulties in autistic subjects on explicit memory tasks, particularly episodic memory tasks. Episodic memory is defined as the memory of personally experienced events, situated in the temporal-spatial context of their acquisition and which implies "mental time travel" back through one's past, associated with autonoetic consciousness. Several studies have argued that there is an elaborate encoding deficit in ASD memory and learning as well as retrieval difficulties especially in free recall tasks while cued recall and recognition are mostly preserved (for review see Desaunay et al., 2020). For example, learning of a repeated local context could slow down processing of other trials thereby limiting the integration of these trails into a new context (Kourkoulou et al., 2012). Other studies investigated the effect of atypical perceptual functioning on explicit memory for complex figures and have revealed large variations in performance (e.g., Prior and Hoffmann, 1990). Results often depended on the index chosen to measure the impact of local processing bias on memory; studies that used an index based on accuracy found significantly impaired performance (Minshew and Goldstein, 2001; Kuschner et al., 2009) with no evidence of a local processing bias (Kuschner et al., 2009). Other studies used the Developmental Scoring System developed by Bernstein and Waber (1996) with four parameters: organization, style, accuracy (with two subscores: the main substructures and the details of the complex figure separately), and errors. Using this detailed scoring procedure these studies found that participants used part-oriented strategies on structural elements suggesting local processing (Schlooz et al., 2006; Tsatsanis et al., 2011).

This part-orientated style has to be considered in a developmental context. Several studies using different scoring systems report a developmental shift from part-oriented to a more configurational style (Kuschner et al., 2009; Tsatsanis et al., 2011). For instance in the Tsatsanis et al.' (2011) study, about 30% young typical children aged from 6 to 14 years preferentially used the part-oriented approach. By contrast, about 10% of the 14–42 years group still used this approach. This percentage contrasts with that of the ASD group where the part-oriented style was present in more than 60% of adults. These results are in accordance with the hypothesis that this "atypical" performance may reflect a delay in development in global/local visual perception in ASD related to maturation of brain connectivity (Kuschner et al., 2009; Crespi, 2013).

Studies of the impact of local processing bias have tended to focus on true memory rates without taking into account false positive errors. Moreover, they have tended to use complex figures as stimuli. These might not be the most appropriate stimuli to address the impact of local processing bias because they provide no data either on intrusions or on false recognitions. By contrast, studies published using verbal learning tasks generate both these measures (Minshew and Goldstein, 1993; Bennetto et al., 1996; Bowler et al., 2000). Other studies have used verbal false memory tasks derived from Roediger and McDermott's paradigm and found contradictory results (Beversdorf et al., 2000; Bowler et al., 2000; Kamio and Toichi, 2007). These contradictions could result from the fact that the impact of processing bias for local information, as described by Frith and Happé, on language is now being debated. This bias would be preferentially observed for non-verbal tasks. Only one used geometric figures with associated visual lures (Hillier et al., 2007). In that study, participants with ASD were better at discriminating true items from false items compared to typically developed comparison participants. We could speculate that if ASD participants had been instructed to process specific details inserted at the same place as the lure items, they would have been more likely than comparison participants to falsely recognize the lures.

The present study aimed to examine the impact of a local bias on both implicit and explicit memory under conditions that promote either local or a global processing. For our implicit memory task, we hypothesize that in the condition favoring local precedence participants with ASD would perform significantly better compared to comparison participants. We predict the reverse pattern in the global condition, where the typical global precedence would favor the comparison participants. In contrast to implicit memory tasks, explicit memory tasks favor conscious mechanisms and globally oriented processing when ASD participants are instructed to focus on the whole of the target. We predicted that in the global condition, participants with ASD would not differ from comparison participants, whilst local processing would increase confusion between targets and lures that share the same details during the recognition test. We tested these hypotheses by conducting two experiments using implicit and explicit memory paradigms. In order to study these effects more in depth and elaborate our cognitive hypothesis, we conducted additional exploratory analyses with more general cognitive functions including local/global precedence, working memory, executive functions, and episodic memory. All these functions were evaluated with standard tests and scores were correlated with those of the experimental tasks.

MATERIALS AND METHODS

Participants

Seventeen participants with ASD and 17 typically developing (TD) comparison participants aged from 10 to 16 years were included in the present study (**Table 1**). The recruitment started prior to the 2013 publication of DSM5, hence participants had all been diagnosed with verbally and intellectually high-functioning autism or Asperger's syndrome according to DSM-IV (American Psychiatric Association, 2000) criteria. The diagnosis was established by experienced professionals using the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994) and/or Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1989). The comparison group was recruited among several French schools. Exclusion criteria for both groups were as follows: history of previous neurological disease (other than ASD in the clinical group), head trauma, current psychoactive medication, intellectual disability, and learning disabilities. Families were given a comprehensive description of the research. Requirements of the local Ethical Committee were met and we obtained written consent from parents of minors, in line with the guidelines of the relevant ethics committees.

General Cognitive Assessment

All the participants underwent a general clinical cognitive assessment including IQ, working memory, executive functions, episodic memory and local/global precedence aimed at documenting their cognitive profile and enabling correlation analyses with experimental memory tasks. Participants' IQ was assessed using the Wechsler Intelligence Scale for Children (WISC-IV, Wechsler, 2005). Groups were matched for age, gender, the Verbal Comprehension Index and the Perceptual Reasoning Index (**Table 1**). The ASD group differed from the comparison group for the two other Wechsler's indices, i.e., Processing Speed Index and Working Memory Index. ASD participants scored poorly on spatial working memory measured with the spatial span task (Farrell Pagulayan et al., 2006). This task is similar to the classical Corsi Block Tapping Task but we calculated a basal score representing the highest level at which the participant correctly reproduced the four sequences. Executive functions were in normal range. These included inhibition (interference score on the Stroop task, Albaret and Migliore, 1999), planning capacities (number of problems correctly solved at the first trial of the Tower of London, Lussier et al., 1998) and strategies of retrieval from semantic memory assessed with two fluency tasks (Cardebat et al., 1990), i.e., semantic (name of animals) and phonemic (words beginning by the letter *p*) fluency tasks. Episodic memory was assessed by means of the story recall task (CMS) and the Rey-Osterrieth Complex Figure Test. ASD participants obtained pathological scores in the story recall task

TABLE 1 | Participant characteristics (means and standard deviations, SD), and analyses for group differences (independent samples *t*-tests).

	ASD (*N* = 17)		Comparison (*N* = 17)		Group differences p and effect size
	Mean	***SD***	**Mean**	***SD***	
Age (in months)	161.87	26.29	161.78	19.99	*ns*, $r = 0.002$
Wechsler Intelligence Scale					
Verbal Comprehension Index	100.50	18.70	111.73	11.01	*ns*, $r = 0.34$
Perceptual Reasoning Index	101.18	14.67	103.93	10.58	*ns*, $r = 0.11$
Processing Speed Index	88.25	16.36	102.27	11.63	$p < 0.05$, $r = 0.45$
Working Memory Index	94.00	16.86	106.60	12.01	$p < 0.05$, $r = 0.40$
Spatial span	4.00	0.63	5.07	1.03	$p < 0.005$, $r = 0.54$
Executive functions					
Inhibition	20.62	9.39	25.20	6.23	*ns*, $r = 0.28$
Planning (1st trial)	7.31	1.35	7.40	1.50	*ns*, $r = 0.03$
Semantic Fluency	29.00	12.80	31.94	5.29	*ns*, $r = 0.15$
Phonemic Fluency	15.12	5.75	17.47	4.31	*ns*, $r = 0.22$
Episodic memory					
Immediate story recall	19.56	11.13	27.80	8.06	$p < 0.05$, $r = 0.40$
Delayed story recall	17.31	11.76	26.47	7.94	$p < 0.05$, $r = 0.42$
Story recognition	12.00	2.42	13.53	1.36	$p < 0.05$, $r = 0.37$
Rey recall	17.78	7.40	21.80	5.29	*ns*, $r = 0.30$
Perceptual bias					
Local precedence	5.28	4.92	1.37	4.69	$p < 0.05$, $r = 0.36$
Global precedence	0.71	3.73	3.51	5.39	*ns*, $r = 0.29$

ns, non significant.

($p < 0.05$). Finally, local and global precedence were investigated with a selective attention task used in the Plaisted et al. (1999) study and adapted from the Navon task (Navon, 1977). Briefly, the participants were presented large letter shapes made up of smaller letters that were either the same as or different from the larger letter. They were asked to process either the large (global condition) or the small letter (local condition) in two sessions where they had to identify either the small letter or the large letter in the presented stimuli. Target letters were "H" and "S." Three kinds of stimuli were provided: compatible stimuli where the large and small letters were the same (S/S and H/H), incompatible where the large letter and the small letter were different (H/S), and neutral stimuli which corresponded to either an "H" or an "S" at a global level when participants had to judge the large letter (a large H made up of small As, and a large S made up of small As) or at a local level when they were required to judge the small letter (a large A made up of small Hs and a large A made up of small Ss, see Plaisted et al., 1999 for methodological details). We calculated two *precedence indices*, one for each condition consisting of the advantage of the compatible trials compared to the neutral. ASD participants differed significantly from the comparison group only for the local precedence index.

The age-related effects on performance were analyzed by means of Pearson correlation coefficients. No significant correlation was obtained in the ASD group. On contrary we observed a significant increase in performance with age in the comparison group for inhibition ($p = 0.001$), planning ($p = 0.05$), and retrieval strategies assed with the semantic verbal fluency task ($p = 0.001$).

EXPERIMENT 1

The first experiment focused on the effect of perceptual bias on implicit memory. The priming task consisted of tachistoscopic identification of drawings of common objects using two distinct encoding conditions: one favoring local processing (Local condition) and the other favoring global processing (Global condition) of drawings.

Participants

All participants took part in this first experiment. For those who participated in both Experiments 1 and 2, the priming task was always conducted first to avoid the interference from the intentional memory strategies on the implicit memory task.

Stimuli

A customized database of 220 colored drawings, divided into 20 semantic categories of living and non-living common objects, was created by a cartoonist. On the basis of a pre-experimental pilot study (200 subjects aged 10–20 years), we selected 160 drawings, which were always successfully identified in under 160 ms, the time limit used in the tachitoscopic task. These 160 items were then divided into 16 lists of 10 items. We ensured that the 10 items of one list belonged to different semantic categories and verified that each list yielded a 50% rate of "yes" responses during the study phase for both local processing (containing a small circle in a particular part of the object) and global processing (the global size of the object that could fit into a square of 10 cm width). To avoid potential item effects, we created 60 combinations, each comprising of six lists of target items shown

at both study and test phases, four lists of distractors which were provided during the study phase only and six lists of control items, provided during the test phase only.

Procedure

Participants were placed in front of a 17-inch laptop screen a in a quiet room. They were shown the different series of drawings using E-prime. The task was divided into a study phase containing both local and global conditions, in counterbalanced order between the subjects, and a test phase with all the studied drawings. The study phase consisted of showing 50 drawings per condition: 30 targets and 20 distractors. Each trial started with a fixation cross placed in the center of the screen for 1000 ms, followed by a drawing presented for 1500 ms and a gray display that disappeared either when the participant responded or when 5000 ms had elapsed. The instructions were different in the Local and the Global conditions. In the Local condition, each drawing contained a pink shape on a small part of the object and the participant had to decide if there was a dot in this shape (**Figure 1**). In the Global condition, an empty frame was provided as a reference measurement to judge if each object drawn was smaller or larger than the frame. Participants were encouraged to process each object globally when performing the task. They responded by pressing one of two keys on a response box. A training phase was provided to ensure that all participants had understood the instructions. During this study, we collected both accuracy scores and response times. After 10 min delay, filled by the Stroop task, the test phase began. This phase was described as a new different task and consisted of a tachitoscopic identification task containing the 60 targets (30 local + 30 global) and 60 new lures, i.e., the non-studied drawings. Each trial started with a fixation cross during 1000 ms followed by a drawing, a scrambled mask specific to each drawing respecting the same perceptual properties as the drawing and designed to limit the persistence of vision and finally, another gray display. The duration of presentation increased until the participant named the object: the first duration was 16ms with an incremental step of 16 ms up to 160 ms maximum. Two breaks of 30 s maximum were introduced during this test phase. The experimenter recorded the number of presentations needed to give the correct answer. As is usual in neuropsychological research of this kind (see Roediger and McDermott, 1993), we calculated a priming index for each experimental condition by subtracting the number of exposures for targets, either local or global, from the number of exposures for new lures. The priming effect is revealed by a significant difference between the two types of items and by a positive index.

Statistical Analyses

Statistical analyses were conducted using Statistica software. We ran analyses of variance (ANOVAs) using a General Linear Model procedure on response time, accuracy and priming scores. We also calculated effect sizes (η^2 or r according to the test used). *Post hoc* multiple comparisons were Tukey-corrected. We also conducted Pearson correlations to test the possible association between age and behavioral performance and the relation between other cognitive functions and priming scores in both groups.

Results

Study Phase

The 2 (Group) × 2 (Condition) repeated measures ANOVA conducted on response time at the study phase revealed a significant effect of Group [$F(1,29) = 4.42$, $p = 0.04$, $\eta_p^2 = 0.13$]. The ASD group was slower than the Comparison group in both conditions (**Figure 1**). No other significant effect or interaction was found (**Figure 2B**).

The 2 (Group) × 2 (Condition) repeated measures ANOVA conducted on accuracy at the study phase revealed a significant effect of Group [$F(1,29) = 5.62$, $p = 0.02$, $\eta_p^2 = 0.16$]. The ASD group performed significantly worse than the Comparison group. We also observed a significant effect of Condition [$F(1,29) = 33.65$, $p < 0.001$, $\eta_p^2 = 0.54$] with no interaction between factors. Performance on the Local condition was higher than on the Global condition in both groups (**Figure 2A**).

Test Phase

The 2 (Group) × 3 (Items: local target, global target, non-studied) repeated measures ANOVA performed on the number of exposures at test revealed a significant effect of Items [$F(2,58) = 32.74$, $p < 0.001$,$\eta_p^2 = 0.53$]. Non-studied items needed more exposures in order to be identified than local ($p < 0.001$) and global targets ($p < 0.001$). Hence, we observed a significant priming effect for local and global conditions (**Figure 3**). In addition, global targets were identified faster than local ones ($p = 0.01$). There was no other significant effect or interaction. However, we noticed that the difference observed between local and global target in the comparison group ($p = 0.04$) was not found in the ASD group ($p = 0.1$).

According to these results, the 2 (Group) × 2 (Priming index) repeated measures ANOVA confirmed the previous results showed a significant effect of priming index [$F(1,29) = 5.98$, $p = 0.02$, $\eta_p^2 = 0.17$] with greater priming in the Global condition compared to the Local condition.

Age-Related Effect on Performances

Pearson analyses revealed no significant correlation between age and any scores except for Global Priming index in the Comparison group ($r = -0.57$, $p = 0.03$) where the magnitude of priming decreased with age.

Relation Between General Cognitive Function and Priming Indices

We conducted exploratory correlational analyses between cognitive functions and the two priming indices calculated for each condition to identify possible contribution of some other specific functions to the priming effects. We obtained some significant correlations but only one survived Bonferroni correction. However, we preferred to report these non-significant correlations after Bonferroni correction (NS) to get an overview of the possible cognitive mechanisms associated with priming in each group. The only significant correlation obtained in the ASD group was the positive association between the global priming index and the phonemic fluency ($r = 0.51$, $p = 0.04$, NS). We observed the reverse association in the Comparison

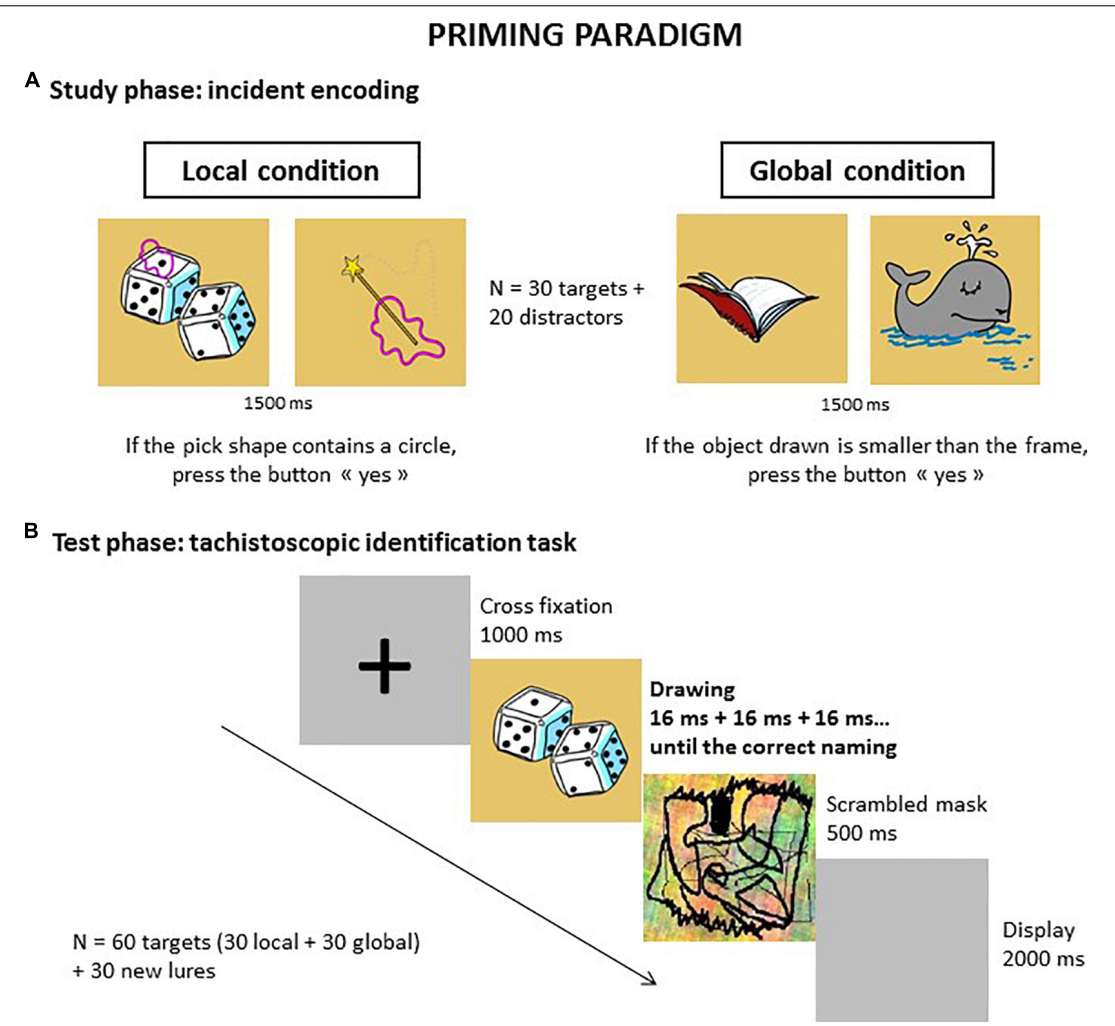

FIGURE 1 | Priming paradigm. The task included a study phase with both local and global conditions provided in a counterbalanced order between subjects **(A)**, followed after a 10 min delay by a test phase **(B)** which consisted in identifying all studied drawings presented with an initial 16 ms time duration and increased with an incremental step of 16 ms up to correct naming.

group: the global priming index was negatively correlated with performance on semantic fluency ($r = -0.87$, $p = 0.03$, NS), planning ($r = -0.75$, $p = 0.001$) and inhibition ($r = -0.50$, $p = 0.05$, NS). Finally, in the Comparison group, only local precedence correlated significantly with the local priming index ($r = -0.56$, $p = 0.04$, NS), where Local Priming increased when local precedence diminished. In other words, this result could reflect the need to go beyond local perception in order to perform the tachitoscopic identification task.

Discussion

The main objective of this experiment was to identify the effect of local or global oriented processing on implicit memory using a tachitoscopic identification task. During the study phase, the ASD participants were less accurate and slower than the comparison group. This slowness is corroborated by the processing speed index derived from the Wechsler test. This characteristic is a well-known feature of autism and is also observed in younger populations (Oliveras-Rentas et al., 2012; Hedvall et al., 2013). Concerning accuracy, participants with ASD made more errors than comparison participants that may reflect greater difficulty making these perceptual judgments. However, the difference in performance between the two experimental conditions is similar to that of the comparison participants. Both groups of participants were less accurate in processing the size of the whole target relative to the standard-sized frame placed beside the computer (Global condition) than they were at processing a dot (Local condition). Beyond these differences in complexity, we observed a significant priming effect in both local and global conditions in both groups. These results are in accordance with previous published priming data collected with different experimental paradigms (Gardiner et al., 2003).

Contrary to our expectation, the analysis of the priming index showed no superiority for the Local condition in the ASD group. Instead, we observed a significant advantage for the Global condition, which was reduced for the ASD participants.

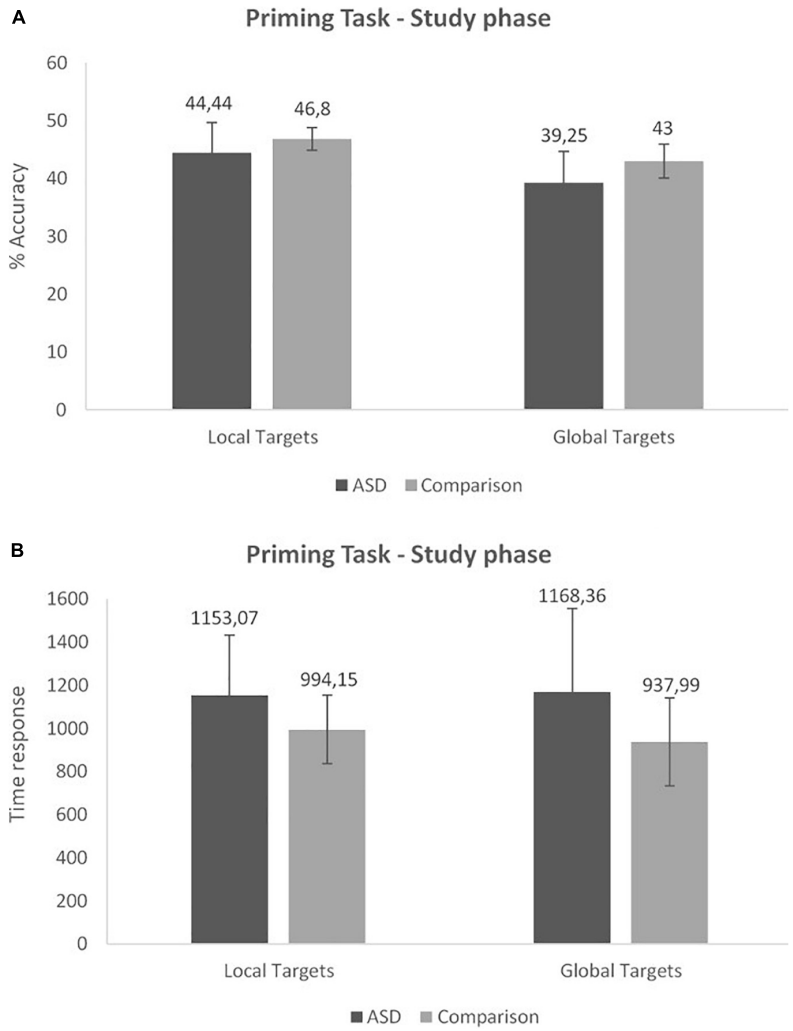

FIGURE 2 | Priming task: accuracy **(A)** and time response **(B)** at the study phase (Mean and SD).

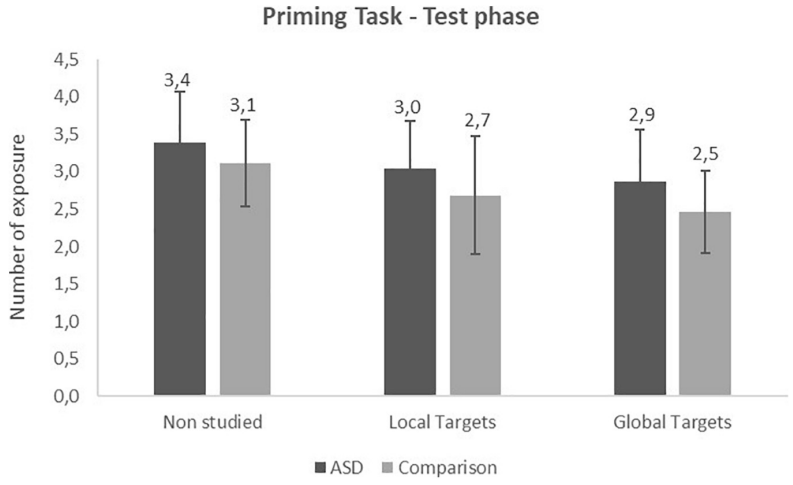

FIGURE 3 | Priming task: number of exposure at the test phase (Mean and SD).

When we considered each group separately, we found this advantage only in the Comparison group. These results confirm our second hypothesis that predicts better performance in the global condition only in the comparison group. This may be because participants with ASD naturally and automatically process details first and consciously extend to the whole drawing in a second step. Note that this improvement remains discrete because local bias may not present be among all ASD participants. The tachitoscopic presentation may constrain ASD participants to a local processing style by not giving them enough time to shift from salient details to the whole shape. This interpretation is in accordance with the results of the correlation analyses. Whereas the control functions tested by the fluency task were implicated in the Global priming of the ASD group, they were not in the Comparison group. In addition, our data showed that local precedence may have had a negative influence on identification performance during tachitoscopic presentation. Taken together, our results suggest that natural perceptual processes oriented toward local information, associated with a lack of attentional disengagement to perform a global oriented processing, in ASD impact upon their implicit memory by preventing global processing in time-limited conditions.

EXPERIMENT 2

This experiment aimed at identifying the impact of the perceptual processing bias on episodic memory. We pursued this question by providing two different recognition tasks of cartoon pictures that favored either local or a global processing. During the "local condition," participants had both to process a specific detail and remember each cartoon followed by a recognition task where targets were mixed with two kinds of lures: half with the same details as those of the target and half with new, different lures. The purpose of this design was to reveal a local memory bias resulting in an increase in confusion between targets and lures when they shared the same details. We expected that this local bias would be more likely in the ASD group. In contrast, the "global condition" consisted of processing each cartoon as a whole by making an indoor/outdoor judgment. As in the previous condition, the recognition task contained two kinds of lures, half with the same background as that of the target and half with new, different lures. An increase in confusion between targets and lures sharing the same background would provide evidence of a global bias in perceptual processing. We expected this pattern to be more evident in the control group.

Participants

Among the children and adolescents who participated in the first experiment, 13 ASD participants and 13 TD participants were tested here [age range: 79–180 months (6.6–15 years), mean = 135.77 ± 28.19 months (11.31 ± 2.35 years)]. All children and adolescents took part in both experiments on the same day. The remaining participants were given another and more difficult version of the episodic memory task that mixed the two conditions, *i.e.,* local and global, in a same test. We observed a floor effect in the first participants, which led us to make the present methodological adjustments.

Stimuli

Two tasks were created; each consisted of 60 cartoon pictures with the same visuo-spatial structure drawn by the same cartoonist as drew the pictures for Experiment 1. The stimuli included an equal number of indoor and outdoor situations. Each task contained a set of 60 cartoons that were divided as follows: 20 targets, 20 specific lures oriented toward either "local" (local condition) or "global" (global condition) properties of targets, and 20 different lures (**Figure 4**).

Procedures

Local Condition

During the intentional encoding phase, participants were shown 20 cartoon pictures, each containing an object surrounded by a pink mark and presented for 2000 ms. The participants were requested to look for a specific detail, i.e., a circle inside the mark, and provide a response as soon as possible by pressing the "yes" or "no" button on a response pad. There was 50% of probability of finding a circle on these 20 targets. They had also to remember the scene. After a 10-min delay, the recognition phase was conducted where the 20 targets were mixed with 40 lures. There were 20 "local" lures that contained the same details located in the same place as the 20 targets, i.e., a "local" lure for each target, and 20 totally different new lures. Participants were asked to discriminate the targets from lures.

Global Condition

This condition followed the same procedure as the local condition. The task was composed of an intentional encoding phase where 20 targets were provided with an indoor/outdoor judgment for each cartoon picture. After 10 min, the recognition phase was conducted with the 20 targets mixed with 20 "global" lures characterized by the same background as the 10 targets (five indoor and five outdoor situations) and 20 other different lures.

For each condition, we calculated three scores: a discrimination index (*d*' or *d*-prime), proportion of Hits, the proportion of false recognitions for specific lures, i.e., local or global, and for the new different lures. The discrimination index takes into account Hits and False Recognitions for specific lures, i.e., "local" for local condition and "global" for global condition.

Statistical Analyses

Statistical analyses were conducted using Statistica software. We performed independent samples *t*-tests and calculated effect sizes (*r*). We also conducted Pearson correlations to test the possible association between age and behavioral performance and the relation between other cognitive functions and episodic scores in both groups.

Results

The ASD group differed significantly from the comparison group only on the *d*' index in the local condition (**Table 2**) with the ASD group scoring lower than the Comparison group ($p < 0.05$; $r = 0.40$).

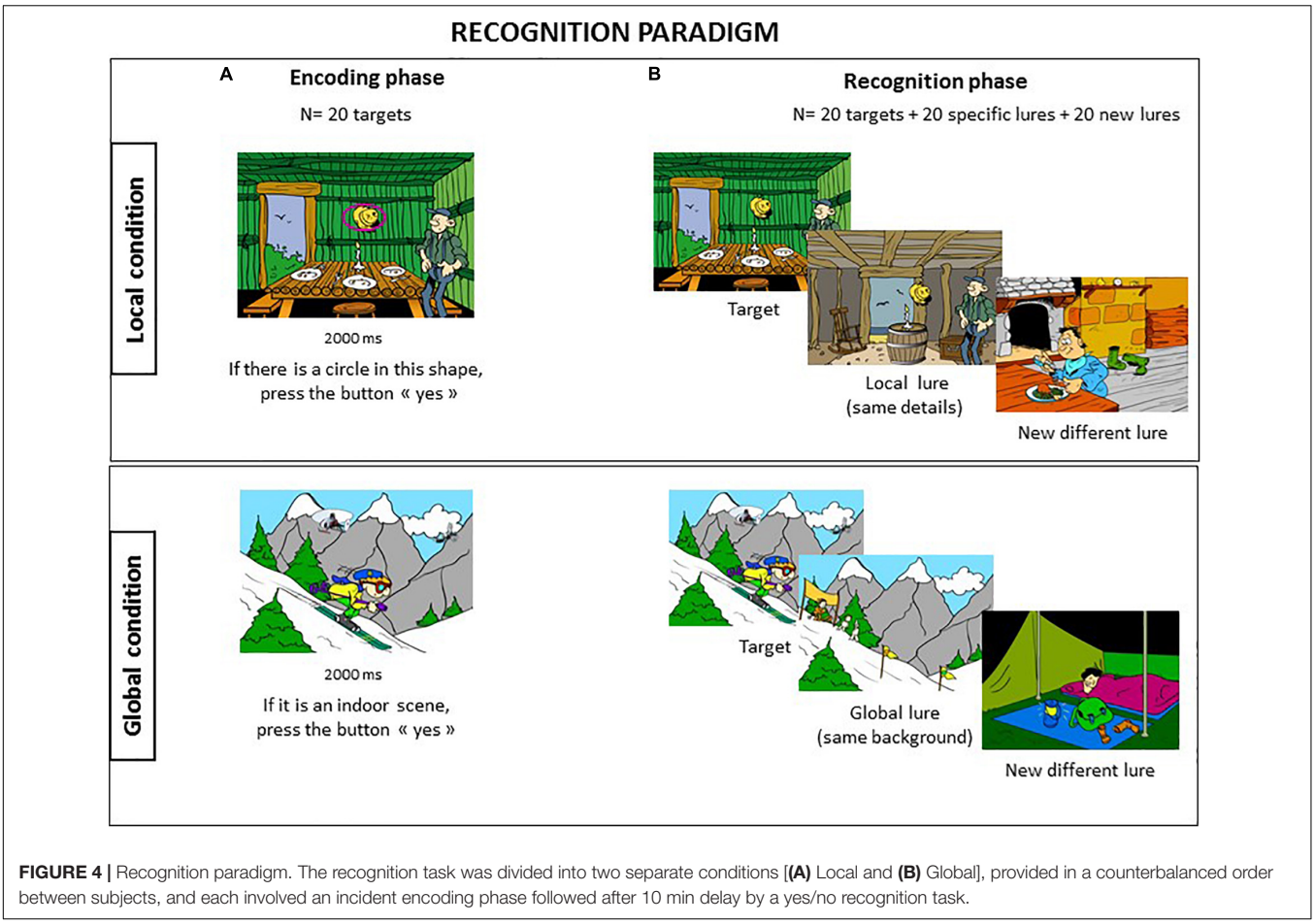

FIGURE 4 | Recognition paradigm. The recognition task was divided into two separate conditions [**(A)** Local and **(B)** Global], provided in a counterbalanced order between subjects, and each involved an incident encoding phase followed after 10 min delay by a yes/no recognition task.

Age-Related Effect on Episodic Memory Scores

Pearson analyses revealed no significant correlations between age and the memory scores (max. $r = -0.44$, $p = 0.17$) except for False Recognitions of local lures in the Comparison group ($r = 0.67$, $p = 0.02$, NS).

Relation Between General Cognitive Function and Episodic Memory

As for Experiment 1, we conducted exploratory correlation analyses between cognitive functions and the discrimination index and local/global false recognitions (**Table 3**). For the ASD group, these analyses revealed significant positive correlations between *d'* in the local condition and the Processing Speed Index, inhibition, and recognition performance in the story recall task. False Recognitions of local lures were negatively correlated with the inhibition process ($p < 0.001$). Other negative correlations were observed in the global condition between False Recognitions of global lures and the Verbal Comprehension Index, planning, and recognition performance in the story recall task ($p = 0.001$). These are interesting but preliminary results that did not survive Bonferroni correction except two of them related to False Recognitions ($p < 0.001$ and $p = 0.001$).

For the comparison group, a significant negative correlation was found between *d'* in the global condition and Rey recall performance ($r = -0.66$, $p = 0.03$, NS).

Discussion

The main objective of this second experiment was to test the possible confusion between targets and lures on the basis of shared local features in participants with ASD. We hypothesized that false recognitions would be higher in the ASD group compared to comparison participants in the Local condition. The data collected confirm this hypothesis by showing that the ASD group had difficulties in discriminating lures from targets when they shared common details. The correlation analysis revealed that these difficulties were associated with processing speed and inhibition. We did not observe any difference between groups in the Global condition but correlation analysis showed that the capacity to reject lures is associated with a more general index of verbal comprehension and recognition memory.

Detail-oriented processing at study reinforces ASD participants' natural preference for local precedence and interferes with recognition judgments. This is not overcome when intentional learning is requested. This result both confirms and extends studies on recognition memory using a recognition

TABLE 2 | Performance at the recognition task.

	ASD		Comparison	
	Mean	***SD***	**Mean**	***SD***
Local condition				
Discrimination index (*d'*)*	0.84	0.58	1.39	0.68
Hits	0.55	0.18	0.62	0.15
False recognitions "local"	0.26	0.14	0.17	0.12
False recognitions "new"	0.08	0.13	0.02	0.04
Global condition				
Discrimination index (*d'*)	2.98	2.51	2.35	1.41
Hits	0.80	0.17	0.79	0.12
False recognitions "global"	0.17	0.23	0.13	0.08
False recognitions "new"	0.12	0.17	0.08	0.10

*$p < 0.05$; $r = 0.40$.

TABLE 3 | Significant correlations (and *p*) between general cognitive function and episodic memory scores in the ASD group.

	ASD			
	Local condition		***Global condition***	
	***d'* index**	**FR "local"**	***d'* index**	**FR "global"**
Wechsler Intelligence Scale				
VCI				−0.62 (0.03)
PRI				
PSI	0.63 (0.03)			
WMI				
Spatial span				
Executive functions				
Inhibition	0.60 (0.04)	−0.87 (0.0001)		
Planning (1st trial)				−0.63 (0.03)
Semantic Fluency	0.64 (0.02)			−0.60 (0.04)
Phonemic Fluency				
Episodic Memory				
Immediate story recall				
Delayed story recall				
Story recognition	0.64 (0.03)			−0.84 (0.001)
Rey recall				
Perceptual bias				
Local precedence				
Global precedence				

VCI, Verbal Comprehension Index; PRI, Perceptual Reasoning Index; PSI, Processing Speed Index; WMI, Working Memory Index; FR, False Recognition.

index that combined correct and false recognitions (Bowler et al., 2000) by limiting this confusion to experimental conditions that promote local processing. Interestingly, this phenomenon seems to be associated with the capacity to inhibit inappropriate but salient details common to target and lure. A relation with effortful or executive functions and memory has been previously reported using relational memory tasks (Maister et al., 2013). In addition, correlation between the discrimination index and processing speed would appear to confirm this hypothesis. In contrast, recognition based on a globally oriented processing is correlated with a more general capacity of verbal comprehension. Picture encoding in memory relies on both verbal and visual coding (Hockley, 2008) and difficulties in verbal comprehension may share common mechanisms with global integration of the scene.

We did not observe the same influence of executive functions within the Comparison group; instead, we found a dissociation between the capacity to recall a complex detailed figure and correct discrimination after global processing. Hence, in typically developing people, global and local memory based processing may be two more independent mechanisms.

GENERAL DISCUSSION

The present pair of experiments investigated the impact of a local bias observed in ASD on both implicit and explicit memory in conditions that promote either local or global processing. Experiment 1 confirmed earlier findings of preserved priming effects in ASD. Overall, both groups of participants showed the same pattern of performance with slight modifications. Our findings showed that participants with ASD seem less advantaged by the Global condition than were the TD group. Experiment 2 focused on explicit memory and revealed a slight but significant difference in the capacity to discriminate lures from targets in the Local condition. Participants with ASD were less able to discriminate targets from lures than were TD participants when the targets and lures shared the same details. Taken as a whole, the data suggest ASD-related difficulties in consciously inhibiting details. Our overall results confirm our hypotheses that the presence of a local bias in ASD may interfere with both implicit and explicit memory processing.

Cognitive Profile of the ASD Group

The two groups of participants were matched on age, gender, verbal comprehension index and perceptual reasoning index. However, the clinical and complementary measures revealed that the ASD group performed significantly worse than the comparison group on processing speed and working memory tasks. The additional assessment of working memory using a spatial span task confirmed this result. These findings are in accordance with the profile published by Mayes and Calhoun (2008). Other findings in the present study suggest that the impairment in long term memory is limited to story recall and recognition. Stories are thought to be instances of complex material that require strategies based on verbal cues in order to understand the whole story. ASD individuals are well known to experience greater difficulties with increasing complexity of the material (Minshew and Goldstein, 2001). The final difference observed between groups concerns local precedence: the ASD group in the present study is characterized by a greater local precedence than TD individuals. The presence of this local bias despite the great variability observed in our groups provides the starting point from which to discuss the impact of local processing bias on the memory performance of our participants.

Impact of Perceptual Bias on Memory

The present study reveals that the part-oriented processing that characterizes ASD may influence both implicit and explicit memory performance. This automatic bias may limit global processing in time-limited conditions as in the present study where tachitoscopic presentations were used. This may have contributed to the overall slowness observed during experimental tasks (Williams et al., 2013) and which is reported by parents in everyday situations that require global processing, and is in line with the arguments of Van der Hallen et al. (2015). In addition, this bias may induce confusion between memories that share common details as demonstrated in the present investigation. This result contrasts with Hillier et al.'s (2007) study which found no difference with a visual paradigm. By contrast, the objective of the task developed for the present study was to test the impact of locally oriented processing on memory using targets and lures that shared strictly similar details for processing during the study phase. Focussing on these details led individuals with ASD to falsely recognize corresponding lures. This result may help to further explain findings from other areas of research, such as Maras and Bowler's finding of lower ASD-related accuracy and higher recall of incorrect details in their eyewitness accounts. Their pattern of results may be a consequence of confusion between incorrect memory and reality when both share similar perceptual details (Maras and Bowler, 2010).

The correlations with executive tasks provide converging evidence for the contribution of attention regulation to memory performance in our ASD participants. First, fluency correlated with priming effects after global processing suggesting that additional attentional strategies may be implicated, enabling participants to go beyond details and process items globally. Second, in the explicit memory task, attentional dysfunction may serve to diminish accuracy in the local condition, i.e., by diminishing correct recognitions and increasing false recognition rates. Once more, when individuals with ASD are instructed to process certain stimulus details, attenuated attention disengagement may prevent the processing of sufficient other information needed to create distinctive memory traces. These data are consistent with current accounts that focus on attentional mechanisms (Kaldy et al., 2013).

There are other findings of the present study that also merit consideration. In both experiments individuals with ASD benefitted from global processing either in implicit or explicit conditions. This advantage may be more limited for implicit and time-limited compared to explicit memory tasks. These results are consistent with the hypothesis that individuals with ASD are able to use global strategies when the experimental design is appropriate (Mottron et al., 2006, 2013b) and when given enough time (Van der Hallen et al., 2015). These considerations raise important issues for interventions. The evidence from the present experiments suggests that if educators were to instruct individuals with ASD to orient their attention toward the whole and to give them more time to process material, they should perform as well as TD individuals. Conversely, orienting attention toward specific details may increase confusion between activities that share similar details.

CONCLUSION

Enhanced perceptual abilities are a well-known clinical feature of ASD. However, there are no studies that have looked at the impact of this superiority for local processing on memory functioning. The major limitation of this work remains the limited size of our sample. However, the present preliminary data bring some arguments in favor of a significant effect of detail focus on both implicit and explicit memory. The study needs to be replicated with a large sample but already highlights the need to take into account atypical perception in some individuals with ASD (i.e., with local bias) in order better to understand memory functioning in a research setting, and to use more specific and appropriate instructions in educational settings.

In addition and given the significant variability that characterizes Autism, it would be interesting to pursue this work to identify individual factors that may influence this pattern of performance.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by ethics committee (CQI) of the French Institute of Health and Medical Research, a methodological committee (CCTIRS) associated with the French Ministry of Higher Education and Research, and the French Data Protection Authority (CNIL). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

KL and BG-G designed the study. JM, LB, AH-D, GM, FG, and EM collected the data. PC participated to the statistical analyses. KL and BG-G wrote the manuscript. DB, FB-B, FE, and J-MB provided substancial modification to the manuscript. All the authors contributed to the article and approved the submitted version.

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Mutual (Mis)understanding: Reframing Autistic Pragmatic "Impairments" Using Relevance Theory

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A central diagnostic and anecdotal feature of **autism** is difficulty with social **communication**. We take the position that communication is a two-way, **intersubjective** phenomenon—as described by the **double empathy problem**—and offer up **relevance theory** (a cognitive account of utterance interpretation) as a means of explaining such communication difficulties. Based on a set of proposed heuristics for successful and rapid interpretation of intended meaning, relevance theory positions communication as contingent on shared—and, importantly, **mutually** recognized—"relevance." Given that autistic and non-autistic people may have sometimes markedly different embodied experiences of the world, we argue that what is most salient to each interlocutor may be mismatched. Relevance theory would predict that where this salient information is not (mutually) recognized or adjusted for, mutual understanding may be more effortful to achieve. This paper presents the findings from a small-scale, linguistic ethnographic study of autistic communication featuring eight core autistic participants. Each core autistic participant engaged in three naturalistic conversations around the topic of loneliness with: (1) a familiar, chosen conversation partner; (2) a non-autistic stranger and (3) an autistic stranger. Relevance theory is utilized as a frame for the linguistic analysis of the interactions. Mutual understanding was unexpectedly high across all types of conversation pairings. In conversations involving two autistic participants, flow, rapport and intersubjective attunement were significantly increased and in three instances, autistic interlocutors appeared to experience improvements in their individual communicative competence contrasted with their other conversations. The findings have the potential to guide future thinking about how, in practical terms, communication between autistic and non-autistic people in both personal and public settings might be improved.

Keywords: autism, intersubjectivity, relevance theory, communication, double empathy problem

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INTRODUCTION

Issues around autistic communication were identified as a top priority for autism research by stakeholders in an independent James Lind Alliance Priority Setting Partnership priority-setting report (Cusack and Sterry, 2016, p. 6). Community priority-setting is an important means of ensuring that research aligns with the needs of stakeholders: something that is essential if we want outcomes to be genuinely meaningful (Milton and Bracher, 2013; Chown et al., 2017). Yet, while language and communication in autism is clearly a key area for research, it remains something of a "blind spot" (De Jaegher, 2013, p. 14; Morrison et al., 2019b): this study addresses this issue. Using a small corpus of transcribed, naturalistic conversations involving eight core autistic adult participants across three different conversation conditions, it explores how implicit expectations of shared relevance contribute to breakdowns in understanding between autistic and non-autistic interlocutors[1].

RESEARCH CONTEXT

Autism

The past three decades have seen interest in autism as a field of research boom [Interagency Autism Coordinating Committee (IACC), 2013; Pellicano, 2014], coinciding with a dramatic shift in terms of how autism is defined (Happé and Frith, 2020). Medically, autism is classified as a neurodevelopmental disorder, hanging on a set of observed and reported behavioral characteristics. These characteristics, largely based on Wing and Gould's "Triad of Impairments" (Wing and Gould, 1979), are described as impairments in social interaction, in (social) imagination (i.e., demonstrating restricted interests and repeated or stereotyped behaviors) and in communication (see DSM-5 criteria, American Psychiatric Association, 2013). Communication, for these diagnostic purposes, "refers to the full range of both verbal/linguistic and non-verbal (including gesture and intonation) means for interacting with others" (Tager-Flusberg, 1999, p. 325).

Autism is also now commonly conceptualized as a form of neurodivergence i.e., "a specific neurological state" (Beardon, 2017, p. 13) or "disposition" (Milton, 2014) that is "different, not less" (Fletcher-Watson and Happé, 2019, p. 23). The study reported on below adopts this perspective. While the shifting parameters and difficulty in identifying a specific biological cause have led to consternation about the validity of the construct that is autism (e.g., Cushing, 2013; Verhoeff, 2013; Timimi and McCabe, 2016), others have argued that the term is nonetheless useful for those whose lived experiences it describes (e.g., Milton, in Milton and Timimi, 2016; Beardon, 2017; Woods et al., 2018; Chapman, 2020).

Based on original findings by Baron-Cohen et al. (1985) and numerous replication studies, autism research has long been characterized by the belief that impaired theory of mind is a defining trait. However, in addition to the recent evidence demonstrating non-autistic people's inability to accurately impute the mental states of autistic people (see below section Autistic Communication and the "Double Empathy Problem"), the idea that non-autistic children and adults consistently perform at ceiling level in ToM tasks has also now been challenged (e.g., see Samson and Apperly, 2010; Warnell and Redcay, 2019). Furthermore, Peterson and Wellman (2019) discovered that autistic children follow a complete, but atypical sequence of ToM stage progression. At the sequential stage when typically developing children are acquiring the ability to represent false beliefs, autistic children are instead developing the ability to understand that underlying emotions can be hidden. It is possible that the over-reliance on false belief test measures in early childhood has skewed our appreciation for the potential of ToM development in autism.

Autistic Communication and the "Double Empathy Problem"

Over the past two decades, research into autistic sociality and communication has begun to turn its gaze toward *intersubjectivity*. Taking a phenomenological perspective, intersubjectivity acknowledges that as embodied social agents we share in some degree of a "co-conception or co-orientation to the world" (Schegloff, 1992, p. 1296). Intersubjectivity functions as a counter to a solipsistic view whereby the individual mind has primacy and emphasizes the inter-relational aspect of selves and selfhood.

Communication, viewed intersubjectively, does not occur in a void, nor solely in the mind of one individual: it is a social and interactive phenomenon. In order to reflect this, and in opposition to traditional explanations of autism that have situated the mind-reading "failures" assumed central to pragmatic breakdown in the minds/brains of the autistic individuals, Milton (2012) proposes the DEP. This holds that cross-dispositional communication (i.e., between two speakers of different neurotypes) is troubled by "a disjuncture in reciprocity between two differently disposed social actors" (Milton, 2012, p. 884), "who hold different norms and expectations of each other" (Milton et al., 2018, p. 1). Misunderstanding or lack of understanding is not a consequence of autistic "impairment" but a mutual failure in reaching consensus through bidirectional empathy.

Recent empirical autism research, situated largely in the social sciences, has begun to provide evidence in support of the DEP and illuminate the difficulties non-autistic people also experience in understanding autistic people: such as difficulty in inferring autistic affective and mental states (Brewer et al., 2016; Edey et al., 2016; Sheppard et al., 2016; Heasman and Gillespie, 2017; Hubbard et al., 2017) and a tendency toward negative thin slice judgements about autistic people (Sasson et al., 2017; Morrison et al., 2019a). Research has also highlighted how autistic people can demonstrate highly successful and nuanced socio-communicative abilities when among others of a similar neurotype (Crompton et al., 2019a,b; Heasman and Gillespie, 2019; Morrison et al., 2019b).

[1]In accordance with the preferences expressed by autistic self-advocates and their allies (see Kenny et al., 2016; Botha et al., 2021), this paper uses "identity-first language" (i.e., *autistic woman*) rather than person-first language (i.e., *a woman with autism*). This choice does not imply a negative judgement toward individuals with autism referring to themselves as such, if this is their wish.

Linguistic ethnographic research (such as that by Ochs and Solomon, 2010; Sirota, 2010; Sterponi and Fasulo, 2010) as well some other work on autistic communication (e.g., Bogdashina, 2005; Chown, 2012; De Jaegher, 2013, 2020; Sterponi and de Kirby, 2016; Di Paolo et al., 2018), has led the way in taking an intersubjective approach to autism and autistic language use. Autistic participants are approached as situated, interactive agents within their familiar worlds, and from "a phenomenological, rather than a biomedical, point of view" (Solomon and Bagatell, 2010, p. 2).

Linguistic analyses that begin with the premise of asking "what is this utterance doing?," instead of automatically problematizing it, can uncover previously overlooked competences. Sterponi and de Kirby (2016) demonstrate that some of the key characteristics of so-called "impaired" autistic language—pronoun atypicality, echolalia and pragmatically atypical utterances—are revealed to have potentially alternative explanations, such as echolalia functioning as a form of perspective-taking. While these studies explore new territory in the analysis of autistic language use, many involve child-adult dyads which are necessarily asymmetric. The present study aims to apply this same approach to an analysis of adult autistic language use.

Monotropism

Monotropism (Murray et al., 2005; Murray, 2018, 2020) is a compelling interest-based account of autism, based within a dynamic, ecological, model of minds. However, it has received little mainstream attention since its conception 15 years ago. Originally proposed by three autistic scholars, the theory begins from the position that the mind is, essentially, an interest system—a starting place not dissimilar to that of the weak central coherence theory—and that "atypical strategies for the allocation of attention" (Murray et al., 2005, p. 139) are the central cause of the various autistic social and behavioral manifestations. Murray et al. propose that the degree or breadth of attention allocation in humans is "normally distributed" and (largely) "genetically determined" (Murray et al., 2005, p. 140), with some people possessing a greater tendency toward multiply focused attention (*polytropism)*, and others a tendency toward more narrowly focused attention (monotropism). Those identified or identifying as autistic will find themselves at the far end of this distribution with a highly narrow "attention tunnel." Where polytropic minds comfortably entertain many simultaneous interests, each moderately aroused, the monotropic mind will maintain only very few simultaneous interests, with each one highly aroused and intensely focused upon.

The monotropic account offers a unified explanation for the many different features associated with autism. The restricted and repetitive behaviors and interests (see DSM-5 criteria, American Psychiatric Association, 2013) can be explained by attention firing into "monotropic superdrive" (Murray et al., 2005, p. 143) and entraining itself onto one self-pleasing task or topic. Crucially, social and communicative difficulties may come about as a consequence of a difficulty in processing, at speed, information from a variety of simultaneous channels (audio, visual, socio-cultural encyclopedic knowledge, etc…); a skill better suited to polytropic individuals with less narrowly and intensely focused attention. Similarly, stimuli outside of the monotropic attention tunnel may carry reduced salience, a potential difficulty when communication is considered in relevance theoretic terms.

Relevance Theory and Mutual Manifestness

Building on Grice's (1975) inferential model of communication, relevance theory (Sperber and Wilson, 1986, 1995) regards communication as involving more than the simple encoding and decoding of a linguistically encoded meaning. Intended meanings are retrieved *via* a context-bound search for optimal relevance, where "relevance" is defined as a balance between the greatest number of communicative effects achieved for the lowest amount of processing effort. The approach is underpinned by two principles. The Cognitive Principle of Relevance holds that the search for relevance is a central goal of human cognition: this is a claim that is backed up by work in cognitive science[2]. The Communicative Principle of Relevance takes it that because human cognition is geared to the search for relevance, speakers ensure that their utterances come with a presumption of their own optimal relevance. Hearers can therefore safely assume that the utterance is relevant enough to merit the effort required to process it. In this way, speakers, therefore, can ensure hearers will pay attention to them.

This mutual calibration of shared cognitive space is central to relevance theory's notion of ostensive-inferential communication. All facts and assumptions both actually and *potentially* available to any individual as a result of interaction between their physical environment and their cognitive abilities are considered "manifest" to them (Sperber and Wilson, 1986, 1995). The set of assumptions that is manifest to an individual at any given time constitutes their "cognitive environment," and two people who share assumptions are said to share a cognitive environment. Finally, any shared cognitive environment in which it is manifest which people share it is described as a "mutual cognitive environment." As Sperber and Wilson put it (1986; 1995, p. 42): "[I]n a mutual cognitive environment, every manifest assumption is… mutually manifest." Mutual manifestness is the basis from which judgements relating to the optimal relevance of an utterance are formed.

For communication to work, meta-representational abilities that enable a speaker or listener to infer what their interlocutor has in mind, and what their interlocutor should reasonably believe them *they* have in mind, are essential. For this reason, relevance theory has largely been used to explain the cognitive mechanisms of (both successful and unsuccessful) utterance interpretation in typically-developed communicators with typical ToM abilities. Of the few studies that have applied a relevance theoretic lens to autistic communication (Happé, 1991, 1993, 1995; Leinonen and Kerbel, 1999; Papp, 2006; Loukusa et al., 2007; Leinonen and Ryder, 2008; Wearing, 2010), all have approached the matter from the perspective that autistic people

[2]That our minds must be economical with what we notice in this vastly information-rich world is now fairly uncontentious. See, e.g., Gigerenzer and Todd (1999) or Clark (2013).

have impaired ToM abilities[3]. Autistic participants have been used as case studies to validate relevance theory's claims on the mechanisms of utterance interpretation.

We suggest that because of their divergent sensory and perceptual experiences (Bogdashina, 2010; De Jaegher, 2013; Beardon, 2017), and markedly different patterns of attention (Murray et al., 2005), it is plausible that autistic people attribute relevance in significantly different ways to non-autistic people. What is and is not relevant, which facts and assumptions are manifest at any given time, and the way in which representations are organized and accessible, may be more markedly different than those of a non-autistic interlocutor, or indeed, a different autistic interlocutor. The degree of cognitive effort required to generate certain cognitive effects will therefore also be different. We argue that both autistic and non-autistic speakers communicate according to the principles of relevance theory. We suggest that it is where assumptions of mutual manifestness are erroneously made (by either or both parties), that mutual understanding will break down. In this way we resituate the responsibility of breakdowns in understanding on the shoulders of all parties involved, as relevance theory has always intended. This position accords with theories that posit that humans are most successful at inferring the mental and affective states of those others who are most cognitively similar to themselves, and that interactions between autistic and non-autistic people are prime examples of where such conditions are infelicitous (De Jaegher, 2013, 2020; Bolis et al., 2017; Fein, 2018; Chapman, 2019; Conway et al., 2019a,b).

MATERIALS AND METHODS

Aims

This study took the form of a small-scale linguistic-ethnographic case-study featuring eight core autistic participants. The primary aim of this study was to investigate the strength of the proposal that the relevance theoretic notion of mutual manifestness might serve to support the DEP-based theory of mutual misunderstanding in cross-dispositional communication, based on an expectation that in such circumstances both interlocutors may be inclined to make faulty assumptions of mutual manifestness.

Participant Selection and Design

Eight core autistic participants were recruited through Assert, a local autism support charity acting as gatekeeper, and invited to take part in three naturalistic conversations of roughly 10 min each. Assert is a member led organization, founded in 2002, that supports autistic people traditionally identified as being "high functioning," or having Asperger's Syndrome, along with their family members, partners or carers. The conversations were focused around the loose topic of loneliness. We wanted to strike the balance between providing some form of framework for the conversations, not unduly directing or influencing their structure, and maintaining a degree of parity across the conversation conditions.

[3]See Leinonen and Ryder (2008) for detailed review.

Since there is a "a lost generation of people who were previously excluded from a diagnosis" (Lai and Baron-Cohen, 2015, p. 1013), and achieving a diagnosis of autism in adulthood is not easy (Taylor and Marrable, 2011), we decided that stipulating a formal autism diagnosis seemed unnecessarily limiting. Instead, autistic participants were asked about their autism diagnosis at recruitment and again within their consent forms. All autistic participants reported a diagnosis of either "autism level 1," "autism spectrum condition," or "Asperger's syndrome:" the various terminology reflecting the differing times at which they received their diagnoses

The sampling in this study was purposeful (Patton, 1999; Palinkas et al., 2015); core autistic participants were selected on account of their being autistic adults who used language as their primary mode of communication as well as their availability and willingness to engage with the research. Within these parameters, we chose to not impose or collect any further demographic stipulations, so as to allow for as much variability as possible. Finding a group of "typical" autistic people is nigh impossible, given the characteristic heterogeneity of autism (e.g., see Beardon, 2017; Fletcher-Watson and Happé, 2019).

Non-autistic participants were asked both at recruitment and within their consent form to confirm that they did not have a history of speech and language difficulties, autism or learning difficulties. Non-familiar stranger participants had been invited to take part in a Linguistics PhD research project looking at communication across pairs of strangers, with no mention made at any stage that their interlocutors would be autistic. The familiar, chosen conversation partners were not asked about an autism diagnosis although in all but two cases they identified themselves as non-autistic, with one chosen partner (Participant code X6) not mentioning it either way, and another (Participant code X3) identifying herself as autistic. The only important criterion for the chosen, familiar participants was the strength of familiarity they had with the core autistic participants.

Making the Experience Meaningful

In order to obtain naturalistic data, it was important to generate and facilitate conversations that were not in any way contrived. In addition, in making the data-collecting activity meaningful in its own right, the research project could become a mutually beneficial endeavor to both us as researchers and to the participants: a cornerstone of participatory and community-based research (Milton and Bracher, 2013; Chown et al., 2017; Elson et al., 2018; Fletcher-Watson et al., 2019).

Loneliness is a "universal affliction" (McGraw, 1995, p. 43) that can not only cause significant distress but also functions as a risk factor for various health problems and increased mortality rates (Holt-Lunstad et al., 2010; Valtorta et al., 2016). Autistic people are especially prone to loneliness and social isolation (National Autistic Society, 2018), further associated with increased depression and anxiety (Mazurek, 2014) and self-harm (Hedley et al., 2018). Given that was potentially relevant to

the participants, we chose loneliness in Brighton and Hove as the central focus of the conversations (see Williams, 2020)[4].

Procedure

Five sessions were scheduled at different times over 3 days in order to make the "Talking Together" project accessible to as many people as possible. In each session, a series of five conversations took place; (1) a core autistic participant (A) with their chosen partner (X); (2) a further A with their chosen X; (3) both core As together; (4) the first A with an unfamiliar, non-autistic participant (B); and (5) the second A with a B participant. The conversations were scheduled for every 20 min, meaning that each core A participant only had one 20-min wait between conversations. Conversations took place in a small private meeting room at the Assert premises in the center of Brighton, just along from the communal waiting room where participants and their familiar partners could wait, talk, rest and have refreshments.

[4]A secondary thematic analysis addressing the qualitative loneliness content—beyond the scope of this primary study—was undertaken and reported on in to ensure that this endeavor was indeed meaningful.

For each of the three conversation pairings, a (different) set of two prompt questions (see **Supplementary Material**) were provided in order to give the participants somewhere to begin, although it was explained that the questions were just there as a guide. Prompts were designed to elicit personal experiences of loneliness, thoughts about loneliness in Brighton and Hove more specifically and to invite ideas around how address those problems within the city.

Conversations were digitally recorded and professionally transcribed according to the transcription conventions adopted for use in Conversation Analysis (originally developed by Jefferson, 1984, see **Supplementary Material**) to include information pertaining to pauses, word stress, and intonation etc., whilst remaining readable.

Data Analysis

Relevance theory is not a methodology but a cognitive theory of utterance interpretation. There is, however, precedent for the application of a relevance theoretic lens to the analysis of conversational data (e.g., Leinonen and Kerbel, 1999; Jagoe, 2012, 2015; Jagoe and Smith, 2016; Jagoe and Wharton, 2021). Jagoe (2015), for example, analyzed the delusional talk of

	Core autistic participant		Conversation condition/configuration	Interlocutor	
	Code	Demographic details		Code	Demographic details
Suite 1	A1	Autistic male with additional learning difficulties, in his 50s	*Cross-dispositional (familiar)	X1	Male work colleague
			*Cross-dispositional (unfamiliar)	B1	Non-autistic stranger, male, early 20s
			*Matched-dispositional (unfamiliar)	A2	Autistic female, mid 30s–mid 40s
	A2	Autistic female, in her mid 30s–mid 40s	Cross dispositional (familiar)	X2	Male friend of A2
			Cross-dispositional (unfamiliar)	B1	Non-autistic stranger, early 20s
Suite 2	A3	Autistic female, French-English bilingual, in her 50s	*Matched-dispositional (familiar)	X3	Autistic female friend of A3's, in her 50s
			Cross-dispositional (unfamiliar)	B2	Female non-autistic stranger, early 20s
			*Matched-dispositional (unfamiliar)	A4	Autistic male, in his 50s
	A4	Autistic male, in his 50s	*Cross dispositional (familiar)	X4	A4's non-autistic wife, 50s
			*Cross-dispositional (unfamiliar)	B3	Female non-autistic stranger, mid 20s
Suite 3	A5	Autistic female, in her mid 30s–40s	Cross dispositional (familiar)	X5	Female Assert staff member, 30s
			*Cross-dispositional (unfamiliar)	B4	Female non-autistic stranger, 30s
			*Matched-dispositional (unfamiliar)	A6	Autistic female, in her 30s
	A6	Autistic female, in her 30s	*Cross dispositional (familiar)	X6	Female friend of A6
			*Cross-dispositional (unfamiliar)	B4	Female non-autistic stranger, 30s
Suite 4	A7	Autistic female, in her early-mid 20s	*Cross dispositional (familiar)	X7	Older sister of A7, late 20s/early 30s
			Cross dispositional (unfamiliar)	B5	Female non-autistic stranger, late 40s
			*Matched-dispositional (unfamiliar)	A8	Autistic male, in his 40s
	A8	Autistic male, in his 40s	*Cross dispositional (familiar)	X8	Female non-autistic housemate and friend of A8
			*Cross-dispositional (unfamiliar)	B6	Male non-autistic stranger, early 20s

Extracts from these conversations are used as illustrative extracts for the purposes of this paper.

seven individuals with schizophrenia engaged in conversation with the researcher (a speech and language therapist) from a relevance theoretic perspective[5]. Relevance theory provided the theoretical descriptive basis for human communication, on which the analysis was built. Furthermore, it served there as the explanatory and theoretical framework underpinning interpretation of the data, with the notion of mutual manifestness (or the lack thereof) functioning "as a useful construct with which to understand the to-and-fro of the meaning negotiation process" (Jagoe, 2015, p. 66). In Leinonen and Kerbel's (1999) relevance theoretic analysis of the talk-in-interaction of three children with pragmatic impairments, transcripts were scanned for "instances of communicative "oddness," created either by the children or the adults" (Leinonen and Kerbel, 1999, p. 372). Approaching the analysis of the data from the theoretical basis of relevance theory, combined with an open-minded, inquisitive attitude and asking "why that, now?" (Sterponi and de Kirby, 2016, p. 398) should, in principle, afford a grounded, reliable yet sensitive reading of the data.

Data Analytic Method

The study presented in this paper uses qualitative methods, situated in an interpretative paradigm. Qualitative coding and analysis is an iterative, reflexive process (Braun and Clarke, 2006, 2020; Tracy, 2010) that develops over an extended period of time. According to Braun and Clarke (2020, p. 6, 7), such inductive and reflexive approaches "fully embrace qualitative research values and the subjective skills the researcher brings to the process." In our case, the analysis took place over a period of months in conversation between the three authors. The primary analysis was undertaken by the lead author (GW—whose doctoral research this research represented) with ongoing supervision, discussion and reviewing of coding, extract selection and analysis provided by the two further co-authors. This triangulation of analytic perspectives, we feel, was further strengthened by our combined diversity of dispositions (two of us are non-autistic and one of us is autistic). The analysts were not blinded as to the autistic "status" of the interlocutors as this would not have aligned with our linguistic ethnographic approach.

In the initial stage of the analysis, the transcripts were read through several times each in order to become familiar with the form and content of the conversations and the individual interlocutors. These first readings were undertaken within the Nvivo data analysis programme (QSR International Pty Ltd, 2020): software designed to assist in the management of qualitative datasets. Some initial codes were made representing emergent themes relating to the loneliness qualitative content, and stored for the planned secondary analysis to be completed later. In those cases where conversational characteristics were already becoming apparent, these were recorded as notes in the research log.

In the second phase of readings, now focused on the primary research aim, printed transcripts were read through, searching specifically for moments of communication breakdown with the view to analyze them through the lens of mutual manifestness. However, it became quickly evident that there were, in fact, very few instances of communication breakdown through the whole 240 min of transcribed conversational data. If anything, these conversations were consistently characterized by sustained mutual understanding. Further discussion of this surprising finding is provided in section Discussion.

The plan was revised to focus instead on the qualitative differences across the different conversational conditions that had become apparent during the note-taking stage in the first readings. Fresh readings were undertaken of the transcripts, this time adopting a "first person perspective" in order or to "bracket out the researcher's own perspectives and assumptions" (Watts, 2014, p. 4). Detailed notes were made on each conversation, capturing observations, impressions, qualities, and patterns. Coding schemes were developed iteratively, guided by the emergent patterns in the data (see **Supplementary Material**). The codes were then organized into four inductively-derived "motifs" (N.B not "themes" as these usually refer to qualitative thematic content): *"flow," "tuning in," "running along the edges of meaning,"* and *"mutual manifestness."*

The *flow* motif relates to instances where conversational progressivity was notably fluid or stilted, as marked by characteristics such as "high-quality turn-taking, short response latencies, and few interruptions" (Koudenburg et al., 2017, p. 51); or pauses (within turns), gaps (between turns) and lapses (between sequences), interruptions and long (monologic) turns.

The *tuning-in motif* brings together characteristics of the conversational form and non-propositional content that indicate that interlocutors are "on the same wavelength" (Koudenburg et al., 2017, p. 53). Features of coordination, such as mirroring the other's speech (either by echoing specific words or phrases or offering parallel anecdotes), and finishing the other's sentences combine with evidence of rapport and the presence of shared jokes and humor (a form of affective coordination: Nelson et al., 2016) to create a sense of dyadic synchrony, or "closely aligned intersubjectivity" (Heasman and Gillespie, 2019, p. 916) that Koudenburg et al. (2017) have termed "emergent we-ness" or "solidarity."

The *running along the edges of meaning* motif borrows its title from an observation made by Sterponi and Fasulo (2010) in their linguistic ethnographic analysis of a young autistic boy ("Aaron") and his mother engaging in verbal play together. Rather than ignoring Aaron's seemingly meaningless utterance playing with the sound of the word "bug," she joins him, echoing his utterances until the sequence develops into a joyful, rhymical duet. "Language [use] is set free and allowed to run along the very edges of meaning" (Sterponi and Fasulo, 2010, p. 135).

There were not many instances of linguistic freestyling, but there were moments of left-field, non-tangential topic development and abrupt topic changes—which echoed the low demand for coherence noted in autistic group interactions by Heasman and Gillespie's (2019)—as well as

[5]N.B. There is certainly no intention to compare autism with schizophrenia, but in terms of communication there are potential parallels in the absence or reduction of mutual manifestness and the consequences that faulty assumptions around this, on the part of both interlocutors, may have.

non-words and word play such onomatopoeia etc. These features all seemed to have in common something of the diverging from ordinary, expected discourse and as such were grouped together under the *running along the edges of meaning* motif.

The smaller *mutual manifestness* motif relates to instances where its presence or absence was clear.

For the final stage of the analysis, the transcripts were analyzed once more: this time from a "third person perspective" (i.e., applying "the analyst's thoroughgoing knowledge of a relevant theoretical and/or substantive literature," Watts, 2014, p. 4). From this stance, extracts that might support, qualify, question or contradict existing literature and the hypotheses driving this study were carefully, purposefully selected and are included below.

Ethical Approval

This study was granted ethical approval by the Tier II Arts and Humanities Ethics Panel at the University of Brighton. All participants were provided with information sheets at recruitment and again on the day of the research and all gave their written, informed consent in accordance with the Declaration of Helsinki. Information sheets were designed with accessibility for autistic people in mind, drawing on GW's personal autistic insights and advice given in the Participatory Autism Research Starter Pack (Pellicano et al., 2017).

RESULTS

The conversations contained very few instances of non-understanding. However, what *was* evident, were discernible qualitative differences between those conversations held by cross-dispositional pairs (i.e., A + X; A + B) and those by the exclusively autistic dyads (i.e., A + A). The codes and resulting *motifs* were developed as a means of trying to capture this difference.

Conversations are presented below in four suites of five (e.g., Suite One includes: **A1** + X1 — familiar cross-dispositional condition; **A1** + B1 — unfamiliar cross-dispositional condition; **A1** + *A2* — unfamiliar matched-dispositional condition; *A2* + X2 — familiar cross dispositional condition; *A2* + B1 — unfamiliar cross-dispositional), so as to allow closer comparison between the three conversations of each core "A" participant. Within each suite, extracts are presented where they are relevant to the primary *motifs* in the following order: (1) *flow*; (2) *tuning-in*; and (3) *running along the edges of meaning*. The first and second *motifs* are closely related to one another and so some extracts may, at times, represent both. For that reason, *flow* and *tuning in* are considered together for each suite. For some suites there may not be extracts representing all three *motifs*. Extracts belonging to the final, smaller *motif* of "mutual manifestness" are woven throughout each suite where appropriate.

Transcripts were organized so that the left column represents the speech of the core autistic participant (A) and the right column their interlocutor (X, B, or another A). Where two As are talking, the As are presented in numerical order (e.g., in Conversation 3, A1 is to the left and A2 is to the right). For readers educated in Western traditions, top-to-bottom and left-to-right biases play a part in how the visually recorded spoken word is engaged with (Ochs, 1979). We wanted to center the voice of the voices of the core autistic participants, even if implicitly.

Suite One

Suite One *Flow* and *Tuning in*

Monologic turns were common in this first suite of conversations. In the cross-dispositional conversation between A1 and X1, A1 appears to stumble over constructing his turns. His speech is peppered with fillers, pauses, stuttered words, and rephrases which means that it takes him extra time to arrive at his intended points.

I felt ↑very lonely.=((sniff)) You know
cos er you know () you know I didn't

have any co↑nnec↓tion_
Yeah
er because of-of erm ((sniff)) erm .h
me living on my own,
You know and er you know de sometimes
I use=I mean I don-w-wouldn't do ↑it
but because of er my loneliness
((sniff))(0.7) I'm not saying I don't
(0.5) ↑FEEL lonely still but it's:ss
not-not-not as ba::ad because erm I've
got the support that I'd-I've got a-a
sup↑port network_

X1, a work colleague of A1's who agreed to come along and participate, is familiar with A1 and appears patient with these long, sometimes labored turns, creating a conversation where A1 has room to speak, but one that has the feel of being lopsided.

In the cross-dispositional conversation between A2 and her familiar interlocutor, X2, there appears to be a greater sense of balance in terms of turn-taking and contributions, but the turns are still often very long (one turn, for example, lasts 45 lines/1 min and 22 s). Again, there are a lot of pauses and gaps, particularly in the first few minutes, and episodes of parallel dialogues where both acknowledge the other's contributions but continue with their own separate topic when the turn passes back to them. Despite A2 introducing X2 as her friend, and them appearing to have a good understanding of each other's day-to-day, the dialogue comes across as rather staid. The conversation remains on a theoretical, intellectual level about the nature and causes of loneliness with not one moment of laughter, enthusiasm, or signal of affect throughout.

In contrast to this is the matched-dispositional conversation, where A1 and A2 meet. Immediately, the conversation has a sense of flow, which continues throughout the interaction. Within

moments of beginning their conversation together, A2 correctly predicts what A1 is aiming for, and helps him get there:

(2.7) I feel lonely (.) ↑BEing there=I
feel on the- I feel'd Bit on the edge_
On-on the edge [y-y-yeah] all the time [Yeah yeah]

Rather than the parallel dialogues of the previous conversation, this one is characterized by a coherent progression of adjacent turns. Where both A1 and A2—most likely for different reasons—had tended toward long turns across the cross-dispositional conversations (familiar and unfamiliar conditions), here they fall into a fluid rhythm of shorter, responsive turn-taking.

Genuine rapport appears to build too, demonstrated by the mirroring of anecdotes and enthusiastic mutual agreement. In the familiar cross-dispositional condition, A2 sat back when her interlocutor (X2) spoke, giving only minimal backchannel cues ("mmm;" "yeah"). Here she seems more engaged, making contributions that could be understood as enthusiastic, further indicating rapport:

They don't even talk to you.
Yeah
You know they think you know what are
you talking [to ↑me for.] [No I think that is troo:oo.]

And you know-d (.) I feel you know
I feel-d er b-ba:ad about that
[You know-d] er you know it's (0.5) [Yeah]
it's a lonely bus journey because

erm I like chatting to people on the
way down-down into town?
↑Yeah (0.5) ↑Yeah no that's true=do you
think that is a British thing?
Oh yeah definitely y-y-y-yeah_
A bit more aloof kind of
=OH yeah definitely y-yeah-yeah I-I-I
would [almost (.) definitely sa:ay] a [Yeah yeah]
British thing.
Yeah.
I-I won't- I-I don't think it's a
European thing but I would say it's
a u-u-European thing.
No it's good?

The shared enthusiasm crescendos around lines 52–101, where they discover they both have dogs. A1's dog is clearly a significant and supportive character in A1's life: he is mentioned in all three of his recorded conversations and also during informal discussions in the waiting room. In this matched-dispositional conversation, mention of the dog appears to spark a long sequence full of laughter, emphatic agreement (e.g., "Me too"-line 56; "YEAH tha-tha-that's why that's exactly what I do"—lines 69–70), shared parallel anecdotes and echoic mirroring of the phrase "love… to bits:"

And I-I-I [love him to bits?] [Never lonely] when you've got a
dog_
=I know I love him [to bits_] [I love my dog
[I absolutely] you know >I was like< to bits too]

The same topic is seemingly met with limited engagement in both of the cross-dispositional interactions. In A1's interaction with his familiar conversation partner the reference to his dog is something of a non-event, although it could be that the dog is already known to his interlocutor (X1) and its mention not especially newsworthy. However, when his pet is introduced to B1—a (non-autistic) stranger to A1—there also appears to be a distinct lack of engagement:

But I-I don't know what SCALE of
loneliness it-it is cos no one's ever
done a .hh a-a e-e cos o-obviously
I've got a do:og.
Mmm.

The focus on the topic of his pet could be framed as evidence of one of the diagnostic features of autism: the presence of "highly restricted, fixated interests" (DSM-5, American Psychiatric Association, 2013). Monotropism theorists, however, have long reframed these intense absorptions—sometimes manifesting as encyclopedic knowledge of a specific subject—as highly aroused interests within a monotropic attention tunnel rather than a cognitive deficit (Murray et al., 2005). In an ethnographic study exploring social interactions at an autistic-separate workplace in Sweden, Rosqvist (2019) identified a mode of engagement she termed "interest-based sociality" that occurred in autistic-only environments:

> [I]interest-based sociality should here be seen as intrinsic group sociality, as a motivator and a driving force for social interaction within a group and a sense of belonging within a community. It includes the importance of having interest-based exchanges with one another, and having common interests and communication based on genuine interest in the topic being discussed. (Rosqvist, 2019, p. 176)

The exchange about his dog in the matched-dispositional condition seems to fit this description. A1 offers up a special interest that is of great importance to him and it is both recognized and reciprocated by A2 who is also passionate about her own dog. It would be tempting to assume some linear correlation between the engaging in a passage of autistically-satisfying interest-based sociality and the ensuing high affect and flow that characterize this conversation. However, the synchrony was already occurring before this episode: a degree of *tuning-in* already appeared to be taking place.

Suite Two

Suite Two *Flow* and *Tuning in*

Suite Two continues with the presence of heavily monologic turns. The familiar matched-dispositional[6] conversation between A3 and her chosen conversation partner X3 (an autistic friend made through Assert), for example, has an opening turn of 37 lines (lasting 1 min and 8 s), peppered only by X3's minimal backchanneling ("mmm, hm mm"). While A3 does tend to dominate the conversational flow (in all three of her conversations), X3 also inclines toward longer turns. At the end of A3's long opening sequence, having invited a response from X3 ("I don't know about you?"), X3 then goes on to hold the floor for a 60-line extended sequence (lasting 1 min 34 s) with just minimal backchanneling from A3.

"Monologues" are one of the examples given under the diagnostic criteria relating to a "failure of normal back and forth conversation" in the DSM-5 (American Psychiatric Association, 2013). While such one-sided verbosity may seem at odds with maintaining conversational flow, in this conversation at least, it does not appear to cause significant disruption. This may because, as McDonnell and Milton (2014, p. 44) have asserted, autistic people "will often feel more in their flow when engaged in monologs or serial monolog style conversations… a practice sometimes engaged in when people on the autism spectrum talk to one another."

Despite the length of each speaker's sequences, the other remains engaged throughout with a sense of rapport, demonstrated by lots of backchannelling, and mutual, enthusiastic agreement. During a passage where X3 is describing how she has found the city much easier to navigate during moments when traffic has been stopped, there is a moment of mirroring of the word "kindness."

	↑then when they then when they stopped
	the tra-traffic for children's parade
[Yeah.]	and [(.)] other parades .h like th-the
Mm.	roads were empty people seemed to be more
	friendly=there-there's a .h there's a
[KINDNESS.]	kind of a .hh [an au↑ra] like an aura
[Yeah yeah]	[au↑ra kind↑ness] and I think it has a
	↑lot to do with (.) traffic ca:rs.

It could be the case that A3 has heard "a kind [of]" (line 56) and wrongly anticipated "kindness" as the coming word. However, while this was not the original word that X3 was working toward, it does seem that A3 has correctly understood the sentiment which is then mirrored back by X3.

The intersubjective synchrony that they appear to share, despite the (on first glance) stiltedness caused by the long turn-taking, is perhaps demonstrated most beautifully at the end of the conversation where they talk about the welcoming, sanctuary-like quality of the café that X3 frequents:

It's really nice isn't it= [ah it's]	[Mmm.]
very friendly and the atmosphere is	
good and you can see the landscape .h	
[so it it's] it's: er () nature so	[↑Ooh yeah.]
obviously it's very healthy sitting.	
	It's very mmm.
I don't know just yeah_	
	Mmm.

In lines 342–344 neither specifies what it is about that café that is of significance or value, or how this somehow functions as a supportive feature toward resilience against loneliness: but they both appear to "get" it. In this moment, whatever that quality of the café might be: it is mutually manifest to both A3 and X3. It is because of this that neither needs to spell it out.

These two speakers appear to be closely attuned. Their monologic turns do not disrupt the flow, perhaps because of the adjacency: both speakers are inclined to take them. The conversation has its own rhythm, its own flow and a sense of symmetry. Progressivity, here, is not rushed; each speaker allows the other to go on whilst maintaining the thread. There is a feeling of natural, structural coordination which may supports the building of "we-ness."

The conversation between A4 and his non-autistic wife, X4 (familiar cross-dispositional condition), presents a very different conversational dynamic. This conversation, for the most part, involves fairly equal, short, and fluid turns. Yet despite this, attunement, rapport, and mutual understanding appear to be low throughout. Unique to this conversation is the proliferation of questions posed to check that they have been understood by, and have properly understood, the other (e.g., "Is that right? Is that what you're saying?"; "Are you talking about…"; "…does that make sense?"). This type of checking-in is often indicative of interlocutors who to wish signal investment in mutual understanding, and demonstrate care and attentiveness. However, combined with moments where A4's attempts at humor seem to fall flat, it might be interpreted as representing two individuals who are struggling to connect. Instead, our interpretation is that it is reflective of the fact that these interlocutors have a long personal history together and have perhaps learnt that in order to understand one another, extra effort must be made. They may know that they often don't understand each other at the first pass and are keen to monitor mutual understanding as conversation progresses.

These speakers, even in these short 10 min of dialogue, describe very different lifeworlds. A4 prefers to spend time by himself, hates parties and struggles to understand what loneliness would feel like. X4 takes pleasure from socializing, likes participating in organized groups and clubs and comes across as very in tune with her own feelings. While it may be the case that they have a lot of shared life experience together, their

[6]This conversational condition was unique to A3 as her chosen interlocutor happened to be autistic, unlike the chosen partners of the other core participants.

subjective experiences of the world—their dispositions—sound very different.

Their apparent difficulty in achieving mutual understanding is epitomized in the extract below where they struggle to understand what the other means, particularly around the definition of "loneliness." A4 has repeatedly been saying that he doesn't "know what the word loneliness means" or what it "feels like"[7]. X4 seems to believe A4 just doesn't experience it as he doesn't need the company of others. From line 222 they fall into trying to define the concept of loneliness. X4 attempts to tell an anecdote describing a moment in which she felt lonely. A4 argues that what she is describing isn't "loneliness." Suddenly the pace changes and where there was a balanced, measured exchange there are now rapid, over-lapping turns:

T- ah that's more like isolated though isn't it?

.hhh O(hhh) or awkward might be not- not be the word lonely.

Yeah see loneliness [it's a] sort of l-lo:::ong term empty [longing you know.] [Yeah I get that-] [Yeah I felt unsettled] at a recent colleague's wedding because you didn't come with me.

And you were like 'ang on I don't [really know anybody here_] [And-and it was only] half my team and they all got £completely£ drunk within five minutes [() ridiculous]=↑no no no but-

[But that's not-]

=That's not loneliness.

This sequence continues with A4 increasingly taking the floor until he interrupts X4 as she begins to respond and more or less continues in monolog form until the time is up, with very little further input from X4. The lack of understanding over what is quite a central issue to this conversation (loneliness), and this inability to synchronize leads not only to a breakdown of mutual understanding but a powerful breakdown of *flow*, and possibly, for this brief moment, rapport.

The unfamiliar matched-dispositional condition (where A3 and A4 meet) seems to have a very different quality. Here again, like in the familiar matched-dispositional interaction, two speakers with the potential for long turns are engaged in conversation, but it seems to *flow* effortlessly from the outset. There is a pace to this conversation, with over-lapping turns that seem to be borne of enthusiastic backchanneling and mirroring of what the other has said, often becoming direct echoing of words or phrases, as demonstrated in the following three short extracts:

NO NO NO not syste[matically] [or possibly]

possibly yeah.

Possibly erm .h the ↑thing is like I

**

=I could work it out.

I w- I can work it out [which never] [Yeah]

**

So: I started #er:rr# looking in other ways to distract myself so (0.5) thank God I had ADHD [kind of] [((Wheezy laughter))] because then I [could KEEP MYSELF] [It entertained you.] () £ENTERTAIN£ MYSELF.

The most striking feature of this conversation, however, and most indicative that these speakers are *tuning-in*, is the immediate and enduring presence of humor and shared laughter, demonstrated in the final extract above. The humor appears to expands a sense of "solidarity" and rapport in which a deeply personal exchange was able to take place (both participants also shared how moving and surprising they had found the experience shortly after recording).

This use of humor to draw an interlocutor into synchrony contrasts with the way in which humor is used by A4 elsewhere. In the unfamiliar cross-dispositional condition (with B3), for example, A4's humor predicts and then undermines B3's earnest attempt to talk about her recent mental health difficulties and the reason she wanted to contribute to these conversations about loneliness:

Mmm.

.hh but erm (.) so I-I hope I can like say something.

M(h) h h h ((squeak snort of laughter)) profound [about] [YEAH] loneliness.

Following this deflective response, B3 ends her attempt to talk about the loneliness she had recently experienced and A4 takes the floor and, whether intentionally or not, this turn acts to maneuver the conversation away from potentially emotive content to a shallower sequence about loneliness facts and statistics.

Humor, then, seems to be utilized in different ways by A4 in these different conversational contexts to achieve different ends. In interaction with non-autistic stranger B3, he appears to be diverting the undesired direction of the conversation, moving it away from the potential intimacy with a stranger. In the familiar cross-dispositional condition (with X4) it comes across as (albeit affectionate) mocking. But it would not be fair to say that A4 consistently employs humor to avoid challenging emotional content, for in the unfamiliar matched-dispositional condition (with A3), humor rises up into natural exuberance, indicative of the spontaneous rapport and in the same conversation he leaves

[7]This kind of response would be typical of an individual with alexithymia: a condition relating to the "difficulty identifying and talking about your own feelings" (Happé and Frith, 2020, p. 10) that frequently co-occurs with autism. The presence of such a condition, particularly if unidentified, would likely contribute considerably to difficulties in mutual understanding.

compassionate space for A3 to weep, and to share some of her childhood trauma:

I had that when I was five yea- like
the worst of the wor:st and I would
I was in agony inside is when I was
and I can picture the little girl on
the STEP_
(2.0) °Aw°
~h~ ((Crying)) ~WATCHing all the KIDS
~ h and WANTing to be part of it (.)
and I never could? ~
(1.0)
~So IT'S [LONELINESS] YOU'VE GOT one [°I'm sorry°]
(2.7) and [aLONE] cannot AND YEAH that [°↑Mmm°]

While A3 finishes her story over the following 10 lines, A4 quietly listens. There is no awkwardness, no attempt to interrupt or disrupt the flow with deflective humor and no stilted pauses when she has finished. Historically, this kind of muted response might have been interpreted as evidence of an autistic lack of interest in the feelings of others. Yet we suggest that this moment does not represent an absence of affective empathy. It is a moment of deep listening: of "daring to go on" (Sterponi and Fasulo, 2010) with A3 and her intimate sharing.

Suite Two *Running Along the Edges of Meaning*

Directly following the extract above, A3 completes her turn by explaining that her coping method, as a young child, was to turn to books. A4 responds by offering his own parallel anecdote, telling A3 how he also read a lot as a child, and used it as a way to access fantasy worlds: "faraway lands and magic and stuff that was all miles and miles away from what was a very isolated childhood I think" (lines 206–209). For someone who has repeatedly expressed uncertainty around the concept of loneliness and what it means for him, this seems an insightful moment. It spurs A3 to share a memory of a book that was special to her, which triggers a creative, playful, exuberant sequence:

buying me this book of erm Noddy Noddy
with er:rm magic eraser=I wanted to
erase the £world£.
[£Be very dangerous£.] [↑↑ha ha ha ha ha↑↑]
.hhh [but ↑then] I felt like I [.hhh Ah::hh]
discovered magic=I had no clue about
it and that was-
=I remember a story about a magic
paintbrush where this guy could pai:nt
whatever he wanted and it would become
real.
↑Yeah ↑yeah [that yeah.] [And it was] like ah so you
could paint a palace you could paint a
[Yeah that's nice.] [stor::m or a dragon] or a-
Sounds lovely.

What makes this sequence so joyful, and powerful, is the fact that they have both dared to play. There is a feeling of an engagement of trust in the other's utterances, scaffolding progressivity out beyond the normal bounds of polite conversation into childlike creativity. They have entered what Sterponi and Fasulo (2010, p. 131) might refer to as a "liminal conversation space." Here, the world—that may, at times, have been experienced as hostile and unwelcoming—can be changed with the flick of a paintbrush or the swish of an eraser.

Suite Three

Suite Three *Flow* and *Tuning in*

Suite Three also features two core participants (A5 and A6) who demonstrate a tendency toward long turns. In the case of A6, her long turns seem to occur as a result of her laboring a little over formulating concise sentences. Like with A1's speech, there are false-starts, fillers, re-phrasings, multiple pauses occasional stutters in her conversation with X6 (familiar cross dispositional condition):

Yeah.
(0.7)
.h I guess I wondered whether or not
jus:t because of the: increase in: .h
(h)a the number of (0.5) people of
that age group living with (.) either
still living with their family or
living with other people of their
age group (.) [.hh] whether or not [Mmm.]
(0.7) I-I mean obviously it didn't
(.) decrease lonelin- have a
significant impact on loneliness but
THAT'S the only thing that kind of
sur↑prised me about it↑ cos I was like
.hhh oh I wondered whether or not that
would have an impact on (.) h
loneliness=but then I suppose the-the
the oger issue is that if you're not
one of those people who's .h living
with (0.2) or-or that I-either those
erm family or friend situations aren't
working out [or] if they are erm .tch [Mmm.]
if they erm (0.7) or if they're just
not in place for whatever reason like
erm .hh er you (1.0) are-are living on
your own for for a different reason
than actually .h other people living

in multiple occupancy houses and
seeming to be .h going out and having
FUN all the time like [(0.2)] erm [Ye:ah_]
might actually increase those feelings
of loneliness.

These features all combine to stall the flow of A6's speech and the pace of the exchange. Where A6 interjects with supportive or enthusiastic backchannelling when her friend (X6) is speaking, X6 tends to sit back when A6 is engaged in formulating a long, sometimes meandering turn. On first glance this may seem like disengagement, but this conversation also seems to feature some moments of affective coordination in the form of shared laughter and cooperative sequences where both parties' turns build toward a shared perspective.

The unfamiliar cross-dispositional conversation 15 (where A6 meets B4) provides a useful comparison. There are just a couple of moments fairly early into the conversation where B4 interjects while A6 is speaking. These interjections are phatic agreements, but because they are more substantial than X6's simple "Mmms" (in familiar cross-dispositional condition) they arguably require more processing effort.

at a table on my ow:n [**I feel really**] [**Yeah I couldn't do that.**]
uncomfortable with?=#Erm# (.) and I
know that some people are really
comfortable with that and like self-
assured enough to do that and stuff .h
and it's just something where I know
that I would be analysing too much (.)
what other people may or may not be
thin↑king **[or wha-what] er::m (0.2)** or **[Ye:::s_]**

On each of these occasions (lines 112–113; line 120), the interjection appears to cause A6 a disruption in her train of thought, triggering a stutter, a filler, a pause. Although the interjection in line 120 ("yes") is only a single word, it is delivered elongated and with flat intonation, marking it as somehow salient and requiring additional processing effort to derive the intended effects (such as an implied attitude or an intention to take the floor). These moments where one is required to simultaneously produce an utterance and process an incoming one can be hard for individuals with a monotropic disposition (i.e., with tightly focused, rather than diffuse, attention). Particularly for those individuals who also have sensory processing difficulties—where parsing speech among a competing cacophony of other (potentially informative) sounds is challenging—a cognitive lag may ensue at moments of high-speed task-switching. These temporary derailments do not seem to affect the potential for rapport. What these two conversations together (both cross-dispositional) perhaps demonstrate is that X6's subdued interjections may be reflective of her familiarity with her friend's need for space when constructing a complex utterance.

A6's second conversation (in the unfamiliar matched-dispositional condition, with A5) begins with a long turn, with no backchanneling from A6 whatsoever until line 26, and then only a handful of backchannels "Mmm"s or "Yeah"s for the remainder of A5's long turn (in total lasting 52 lines/1 minute and 44 seconds). Ordinarily this might indicate minimal engagement. In the context of A6 potentially requiring more time to process linguistic inputs (as discussed above), it might be tempting to wonder whether she is taking time to acclimatize to the language use of a novel interlocutor. Yet A6 begins her first turn (in line 53) by answering with a series of short responses, almost list-like, in response to the points A5 has made. It is here (lines 53–86) that the pace begins to pick up with A5 acknowledging each of A6's comments enthusiastically, creating what might be described as a conversational volley.

Perhaps it is the momentum that has been building that sets the stage for synchrony, but in the following sequence the pair arrive at a moment of mutual understanding—of mutual manifestness—around the meta-perspective-taking of an imagined other:

Yeah I-I've wondered that as well er
=like I remember when my early
twenties my mum was really concerned
about me that I didn't have enough
friends as she [saw] it=she was like [Mmm.]
oh you needed to .h you need to make
more friends and-and like .h that kind
of thing and ()-
GET TO THE RIGHT NUMBER_
Yeah-yeah apparently so.
M hm hm

Here we assume that this hypothetical "other" (based, initially, on A5's mum) is non-autistic, and this is where the niche of this particular moment of mutual manifestness works. In this moment the othering routinely experienced by autistic people is flipped, and A6's "GET TO THE RIGHT NUMBER" is an echoic parodying of an imagined non-autistic perspective. A shared in-joke is created, based on the shared and unifying experience of being judged by an external "normative" perspective that both speakers can (a) speak to and (b) safely assume their interlocutor, being autistic, is also familiar with.

From here the conversation flows into a dense sequence of apparent close attunement with overlapping turns where they are not so much echoing each other as speaking in sync:

Whereas I see it as my choi:ce but is
it (.) my choice because .h this is
now what I do?
Yeah.
=Er-er::r like a (.) as you say
coping strategy or like just a .h a
w- a way [of .h yeah.] [Or even as you] said with
[Yeah.] the theatre tickets [it's] BETTER than
[**vulnerable** putting yourself in a [**vulnerable**
position yeah] **position** where] you might be
[**pointed**] disap[**pointed**] by other people.
=YEAH exactly [yeah yeah_] [yeah yeah_]

Most distinctive about this next phase of the conversation, however, is the dramatic shift in fluency of A6's speech. The stumbles, the re-starts and the drifting, long utterances are almost immediately eradicated and in their place, there is a concise, assured voice:

	°It's interesting.°
Erm er so yeah I don't know [°er	[BUT IT'S
yeah°_]	GOOD that] you say that you've got more
[Yeah.]	friends now [cos you] obviously (0.5) are
	able to (0.2) accept people as your
	friends.
	**
Yeah massively [so.]	[Erm] but THEN AGAIN (1.5)
	I guess that's the thing .h you use all
	those social things as a kind of a
	PLATform to meet other people and
[Yeah.]	[hopefully] .h conVERT that into
[Yeah yeah_] Exactly_	relationships [of some] kind.
	.h erm but it doesn't have to be that
	way_

One possible explanation for this increase of flow of A6's own speech is that this is now her second conversation so she has had time to shake off any initial nerves associated with being recorded. However, as we saw above, in the subsequent unfamiliar cross-dispositional condition, she reverted to the earlier lack of fluency.

The rapport, flow, and attunement (in the form of backchanneling and agreement), remain until this conversation closes shortly after, as does A6's new-found ease of expression. This high level of rapport, attunement, and flow had not appeared to be present in A5's earlier conversation either. The familiar cross-dispositional conversation (between A5 and X5) seemed to lack flow, perhaps on account of the protracted monologic turns taken predominantly by A5. While some rapport was present (evidenced by moments of occasional phatic laughter, and consistent backchannelling throughout), it remained restrained.

In A5's final conversation, in the unfamiliar cross-dispositional condition, she meets non-autistic stranger B4. In contrast to the long turns with her familiar non-autistic conversation partner X5, it begins with a smooth sequence of shorter, interactive turns that flows easily, perhaps because she has come directly from the highly fluid matched-dispositional conversation with A6. Similar to the kind of subjective differences seen in the conversation between A4 and his wife X4, these two speakers describe very different lifeworlds. B4 likes "going out," to the pub or to gigs and ideally in large groups. In contrast, she has had to work hard not to feel self-conscious being seen alone in public places (like a café). A5 tends to do things on her own. Yet this pair acknowledged and approached their differences with a kind of warm curiosity. They ask questions of each other: not "have you understood me?" but "tell me more…":

yeah so does that is that because you	
<didn't enjoy> u͡ni [or:: when you	[I really enjoyed it.]
got-]	
	**
.h so where did you find your tri:be	
(0.2) since you've been a student=is	
	**
Really yeah_	
	But have you always had that sort of
	(0.5) you've never like had that hang-up

Very early on in the conversation, B4 shares the observation that her experience of being a student was quite lonely. As she put it, she had not been able to "find her tribe"[8]. In offering up this information, B4 exposes a degree of vulnerability from the outset. Considering these interlocutors are strangers, this is quite a bold move and one that invites intersubjective alignment and rapport. More than that—and not necessarily knowing that this might be the case—it sets the scene for common ground. While it is not expressed directly by A5 that she too experienced difficulty a community with whom she could connect, it is a common theme of autistic experiences.

> By the time we reach adulthood, autistic people's experience of "togetherness" has likely consisted of some combination of: being intruded on by other people wanting us to engage with them, when we don't share that desire; being interested and curious about other people, but finding them confusing and overwhelming to be around; trying to engage with other people, and having frustrating and unsuccessful encounters; managing to engage "successfully" with other people, and finding ourselves drained and possibly even damaged as a result of what we had to do to "succeed." (Sinclair, 2010, para. 3)

Here they appear to have inadvertently arrived at a means of bridging two mismatched dispositions: by naming, early on, a feeling of un-belonging that it is likely they both can recognize. Despite this conversation being both with an unfamiliar interlocutor and in the cross-dispositional condition, there is a far greater sense of *tuning-in*, as compared to the earlier conversation between A5 and X5 (familiar cross-dispositional). With its rapport, affect, and synchrony it seems to establish a sense of we-ness that might serve as a temporary community, with all the nourishment that that might bring.

Suite Four

Suite Four *Flow* and *Tuning in*

This final suite begins with a conversation between A7 and her elder sister (familiar cross-dispositional). Unlike many of the other core autistic participants, A7 does not dominate the floor with long turns: if anything the conversation is guided by X7 as she poses the questions and ventures points to discuss. The conversation lacks much enthusiasm or "spark" and, listening to the recording, both participants speak in low, quite hushed tones

[8]We recognize that this term can be problematic, and potentially culturally appropriative but we wished to remain true to the participant's words and the spirit with which they were uttered.

with a consistently flat intonation. In addition to the frequent cross-talk there are regular gaps and lapses.

However, both interlocutors seem keen to engage with the other and progress the conversation. They each contribute and respond relevantly to each other's utterances. Yet despite this, the conversation appears to flow like two strangers trying to dance and repeatedly, apologetically, treading on each other's toes:

[It's weird.]	Yeah (0.5) n-[and I wonder] if like social media has more of an impact on it
[Yeah I was sort of thinking that.]	now [(0.5) than in the pa:st,]
(0.7)	
[That's why] we don't think it cos you get the façade of social media making it look like your having an amazing time.	Cos I guess [the-the]
	Oh definitely yeah_
I suppose everyone uses that to hide stuff °°in a way_°°	

This lack of *flow* also seems to corresponds with an absence of *tuning-in*, perhaps because, as we saw in the cross-dispositional condition involving A4 and his familiar partner, these two speakers have quite different life experiences and lifeworlds despite being sisters. X7 has settled into married life and lives with her husband and two very young children. She, too, grew up in Brighton and now often bumps into old and new friends when she's walking around. A7 lives in a shared house, has very few friends and in spite of trying hard to meet people in organized social activities ("meet-ups"), finds it hard to make meaningful connections.

A7 has been explaining that she not only finds it hard to meet people she can connect with in Brighton, but that the fact she grew up locally makes her feel more self-conscious about not having many friends here ("I feel like the weird one for being, like, I've actually grown up here. I've lived here most of my life but I'm lacking people even though I'm in Brighton"). X7 is making attempts to console A7, telling her that this lack of connection A7 is describing is really due to chance (and perhaps attempting to imply that it is therefore not attributable to anything intrinsic to A7).

↑No because that's just circumstances they've moved aw:ay.	
(1.5)	
YEAH I suppose that's true (1.5) I'm just trying to say it without [it °being like°]	
	[Don't wanna] admit it almost_ (1.2) or that you do sort of () talking about it but
Am I.	
	↑Well yeah (1.7) MAYbe uh maybe you don't talk up as much all the time_
	(3.0)
	But you do let us know when you're feeling lonely I would say?
Yeah (1.0) I just also try not to (0.5) do it too often [(1.0)] that's the whole I always say I feel like a burden so I don't do it () [I do] but I don't all the time.	
	[Yeah_]
	[I KNOW it's]
	Yeah and that's difficult because (1.0) I've got (0.2) my family as well so
	(0.5)
Yeah.	
	Obviously you're part of my fa£mily h but you know what I mean£.

A7 seems to be trying to express a sense of isolation and alienation from the wider society that can be a common experience for autistic people. Inadvertently, in trying to comfort A7, X7 may in fact be undermining A7's attempt to share her pain. This moment of missed mutual understanding continues as each continue to talk from their own conflicting perspectives.

The lack of mutual understanding does not appear to stem from a lack of desire to connect. Here are two sisters who appear to care for each other a great deal but their dispositional difference, in this conversation, is seeming hard to bridge. While, in the above lines, A7 seems to be trying to voice a profound loneliness and a sense of not knowing how to reach out, X7 maintains the belief that A7 always lets them know when she is feeling lonely. What else can A7 really say other than "yeah…" (line 305). As the conversation draws to a close, and following X7's suggestion that A7 should send a text or even call someone if she felt really lonely, A7 tries one more time to make her sense of detachment from others around her understood:

I just never know if anyone will answer.	
	Yeah_
	(1.7)
	That is (0.5) the problem with phones as well these days isn't it it's like (0.5)

These speakers appear to be talking at cross-purposes. A7 is, seemingly, trying to talk about the unreliability of people while X7 is talking about the unreliability of modern technology. The information that A7 is working hard to convey is not mutually manifest here, leading to a breakdown in mutual understanding.

This all contrasts with the way in which the conversation involving A7 and B5 unfolds (the unfamiliar cross-dispositional condition). As with all the non-autistic stranger participants ("B"s), B5 does not know that A7 is autistic. Unlike the familiar cross-dispositional condition, where cross-talk was prevalent, here there is none. Turns are well-balanced, representing a

consistently fluid back-and-forth. While A7 still has pauses mid-speech—where she appears to be preparing the next part of her utterance—there are very few of the lapses and gaps that punctuated the earlier conversation with her sister.

For A5 and B4 in the same conversational condition (unfamiliar cross-dispositional), the discovery that they had both experienced difficulty in finding a community they could belong to, opened up space for shared solidarity. In the same way, A7 and B5 also find several things in common, such as the invisibility to others of their deep loneliness and an aversion to socializing in a context fueled by recreational drugs (something they describe as common in the local social scenes). Around lines 78–89, B5 shares the observation that for her, one of the challenges of approaching new people is the fact that it's hard to know for sure whether they are a "good person" or not. Although A7 does not volunteer any further contribution to this topic, she does agree emphatically:

nervous [quite often_] [There's also] approaching
people that er::m (0.5) er-how do you
know if they were a .h good person.
YEAH.

So-called "social naivety" has long been associated with autism (Lai and Baron-Cohen, 2015), and instances of interpersonal victimization (or "mate crime") are unfortunately common among autistic people (Pearson et al., 2020). Whether or not A7 has had direct experience of this herself, she is likely to be at least aware of the potentially increased risks.

Finally, a further similarity between the unfamiliar cross-dispositional conversations of A5 and A7 respectively, is the way in which the opportunity for rapport and intersubjective alignment has been created by the sharing of some personal information by one of the speakers. B5, for example, talks about not having had a family growing up and how, now, it means that she doesn't "have people that I could just go to that just accept me and will listen to me" (lines 210–212). The sentiment expressed here sounds very similar to A7's "I just never know if anyone will answer" in her familiar cross-dispositional conversation [and, incidentally, echoes A1's "when you phone it (the mental health helpline) no one ever answers"]. It is the "daring to go on" (Sterponi and Fasulo, 2010) by making some private aspect of the self-visible, that invites the possibility for mutual understanding on a deep level.

Suite Four *Running Along the Edges of Meaning*

In the conversations involving A8 there is a distinct lack of *flow,* although the extent to which *flow* is disrupted varies between conversations. There is something idiosyncratic about A8's speech that sometimes can make it challenging to parse as a reader: but in the real-time back and forth of each conversation his interlocutors do not appear to notice directly. Structurally, A8's speech can jump at times between propositions that are not clearly coherent, but the difficulties occur most frequently at (and sometimes within) the level of a single word.

The precise nature of these errors is not clear from the speech sample available, and we had no access to any detailed assessment of speech and language, nor know whether this participant has ever had contact with speech and language therapy services. The errors may represent a developmental pattern of a speech sound disorder which A8 could have had since childhood. Speech sound disorders, while under-investigated in autistic people, have received increasing attention (see Wolk et al., 2016). Equally possible, however, are that these errors may be "paraphasias:" the term given to the presence of errors in an individual's speech, sometimes as the use of wrong words ("verbal paraphasia"), sometimes as wrong or switched phonemes ("phonemic paraphasia") or sometimes as half-correct words ("neologistic paraphasia:" Millea, 2013). Although far less discussed than, say echolalia, paraphasia is also associated with autism, and there appear to be instances scattered among A8's speech (e.g., "everything" for anything, and "seeper" for cheaper, Conversation 18, lines 67 and 135; "meed" for mean, Conversation 20, line 68; and numerous verbal paraphasias)[9].

Seen on the page these instances of word-level differences may jump out as odd or disruptive. Yet most of them are easily interpreted within the context of the surrounding utterance. At high-speed, given that a listener is already predicting what will be said before it has been spoken (Kikuchi et al., 2017), they may easily have gone unnoticed. It is possible, however, that they do contribute to the general stiltedness that colors these three conversations, not least because the occasional re-starts and re-phrases indicate that A8 is, to some extent, aware of these mis-speaks and attempting to monitor them. To do this, whilst also following his interlocutor's speech and crafting his own responses, is likely to add to the cognitive demand. It is little surprise that this might entail extra processing time in the form of pauses, gaps, and lapses.

A8's first conversation, in the familiar cross-dispositional condition with X8, lacks flow; there are a lot of gaps and lapses, frequent topic changes, and seemingly missed opportunities to extend or directly respond to what the other has said. Overall there is a sense of rhythmic awkwardness, as if both of them wish to keep the conversational ball in the air, but are finding it difficult to do so. For example, early in the conversation X8 shares an anecdote from when she had been walking recently in the countryside and was greeted by a stranger. A8 attempts a parallel response about how similar things happen when he goes for a walk near where his parents live, but stumbles a little and his response lasts just three lines ("Yeah cos with my parents are they… you they… you know if you…go on a walk… there…most people say hello"). There is a short lapse, then A8 re-takes the floor ("But going back to London…"). He proceeds to comment on something he has heard about London lacking racial integration, but it comes out awkwardly:

> "People with the same backgrounds stay together so like, whites would stay together and Asians would stick together and all that. There's no, like, I could be wrong but there's no re-interaction between mixed races…"

The sometimes abrupt topic shifts between turns seen in this conversation give the impression of two parallel dialogues

[9]Here the word "wrong" does not connote any negative judgement.

maintained over several turns. This dynamic is far more pronounced in the unfamiliar cross-dispositional condition where A8 meets non-autistic stranger B6. Unusually for these conversations, it is not the autistic participant (here, A8) taking long, monologic turns but B6. From the outset, B6 seems to dominate the conversational flow; his first turn is 50 lines long (lasting 2 min and 4 s), interjected only by one "Mmm" in line 35. This becomes a pattern during B6's long turns, where A8 provides minimal backchannel support but does not direct the conversation. It seems possible that A8 lets B6 run on because he is not entirely following B6's points. In his other conversations (familiar cross-dispositional and unfamiliar matched-dispositional), A8 tends to interject yet during B6's extended opening turn, A8 does not make use of many and ample pauses mid-flow. When he eventually re-enters the conversation (line 55), he initiates a new topic where he explains how long he has lived in Brighton and who he knows here, punctuated by several pauses. He then acknowledges B6's previous contribution ("…but it's a, it's an interesting point what you made, erm"), but picks out the incidental mention of the word "London" from much earlier in the conversation, rather than the B6's most recent point that he has experienced a lot of loneliness while being at university ("…but it's a, it's an interesting point what you made, erm, I mean the London, I don't go to London that often but I, they don't speak to each other on the tube they just listen to music").

In the moments throughout the rest of the conversation with B6, when A8 does step in and take the floor, it appears to be to re-orientate the discussion back to a question related to the prompt cards (e.g., thinking about potential solutions to loneliness locally). In the same way that, in the familiar cross-dispositional condition, A8 and X8 would acknowledge each other's contributions but attempt to pursue a new direction, this conversation only just hangs together in terms of coherence.

In the unfamiliar matched-dispositional condition, where A7 and A8 come together, the conversation seems to have a more stable central point of gravity than A8's other two conversations. There is a symmetry in turn-taking and progressivity of the conversation and despite the still-present gaps, pauses, and lapses on the part of both speakers, this conversation nevertheless seems to *flow*. The conversation begins with the pair cooperating, *via* a series of short turns, to establish a joint definition of "loneliness:"

	Mm-what does loneliness m- (.) mean to
	you,
Erm (0.5) suppose it's actually quite	
hard to defi:ne (0.2) but like (0.5)	
	M-I'm guessing like (0.5) on your ow:n no
	frien:ds_
Yeah feeling like you don't have other	
people_	
	Ye:ah.
Kind of (.)to support you °or° (0.7)	
[yeah] feeling actually alone.	[YEAH.]
	Ye:ah.
	(2.2)

A8 poses some questions for A7 ("have you ever experienced loneliness in Brighton and Hove at all;" "do you know people or can you talk to people here?") that, although they are perhaps a little stilted and led by the prompts, remain relevant and cohesive with the previous turns. A little later, A8 shifts topic again, asking A7 whether she thinks things like meet-ups might help to address loneliness in Brighton and Hove (a topic that he attempts to raise again in the subsequent conversation with B6, to no avail). Fortuitously, A7 has some experiences with meet-ups, as she described in the earlier (familiar cross-dispositional) conversation with X7. This triggers a fluid exchange that continues across 101 lines and 17 turns (and lasting 2 min and 28 s) divided across both speakers. This passage evolves naturally from meet-ups, to the time and money required to do them, to the working hours they both have, to how work in various sectors impacts on the ability to socialize. It is perhaps significant that the discovering of a shared interest initiated this extended, fluid passage of interaction.

What marks this conversation out from the other two in which A8 participates, is the fact that he appears able to sustain focus and coherence for far longer stretches. Moreover, his contributions are more directly relevant. While the enthusiastic rapport that we have seen in some of the other pairings seems to be lacking here, so too is the sense of awkwardness that is sometimes present in both the cross-dispositional conditions (familiar, with X8 and unfamiliar with B6). It is difficult to assess exactly what it is that makes this matched-dispositional conversation with an unfamiliar person function more successfully. There could simply be some degree of luck in A8 introducing a topic (meet-ups) that has some resonance with A7. Given that the other topic-related sequences also run on though, there is probably something else occurring here too. In their study investigating neurodivergent intersubjectivity, Heasman and Gillespie (2019, p. 910) found that conversations involving only autistic interlocutors had "a low demand for coordination that ameliorated many challenges associated with disruptive turns." It may be that in this matched-dispositional condition there is implicitly less pressure for A8 to provide highly contingent contributions at all times and that this, ironically, allows him the space to provide them.

DISCUSSION

This study sought to investigate how implicit expectations of shared relevance contribute to breakdowns in understanding between autistic and non-autistic interlocutors. Eight core autistic participants engaged in three short conversations about loneliness: with a chosen, familiar conversation partner ("X"), with an autistic stranger ("A") and with a non-autistic stranger ("B"). Mutual understanding was unexpectedly abundant during these conversations across all types of conversation pairings.

Clear patterns emerge when the four Suites of conversations are considered together. The most striking of these is the difference between conversations that involved two autistic participants (i.e., the matched-dispositional conversations) and

those that involved cross-dispositional pairs. All five matched-dispositional conversations seem to be characterized by a significant (and sometimes dramatic) increase in flow, rapport, and intersubjective attunement. Conversations 3, 6, and 8 are colored brightly by enthusiasm and mutual affect. In contrast, all but a few of the conversations with non-autistic participants lack the above, even when interlocutors were well-known—and had been for a long time—to the core autistic participant.

The fact that interlocutors built rapport, flow, and synchrony far more effectively when both parties were autistic, even when they were strangers, seems to support theories that suggest we get on best with people who have similar minds (De Jaegher, 2013; Bolis et al., 2017; Fein, 2018; Chapman, 2019; Conway et al., 2019a,b). This, in turn, adds to evidence that counters the ToM-deficit theory of autism and bears out anecdotal evidence from autistic people that they sometimes find barriers to social communication minimized when engaging with other autistic people. For example, autistic academic Sinclair (2010, para. 42) observes that "the 'same planet' metaphor, along with metaphors about 'speaking the same language' or "belonging to the same tribe" are very common descriptions used by autistic people" who have had the opportunity to experience an autistic-dominant space. Similarly, autistic participants reported finding matched-dispositional interaction (i.e., with other autistic people) much more comfortable, in a study by Crompton et al. (2019a). Finally, while it perhaps shouldn't need to be said, the very presence of the high rapport and mutual interest demonstrated in these conversations contributes to the literature that challenges the reduced social motivation hypothesis of autism (Chevallier et al., 2012).

One further pattern is that some autistic participants (A1, A6, and A8) appeared to experience optimal individual communicative competence when engaged in exclusively autistic dyadic conversations. For example, A1's turns are shorter and similarly more coherent in the matched-dispositional conversation compared to the cross-dispositional interactions, contributing to a fluid progression of adjacent turns as opposed to the parallel dialogue of his previous conversation in his familiar cross-dispositional conversation condition. Similarly, when talking with A5, A6 is dramatically more fluent. Stumbles, pauses and re-starts that characterize the typically long utterances of the other two conversations are almost entirely absent and replaced with concise, cogent turns. Conversation 19 is the only one of three where A8 was able to maintain prolonged sequences of engaged, coherent turns.

This finding potentially lends support to a monotropic theory of autistic cognitive processing, explained by relevance theory. In those circumstances where increased mutual manifestness makes understanding less effortful (in both a technical relevance theoretic, and an intuitive sense), more cognitive resources are available for language production. Furthermore, according to the theory of monotropism, the attention of monotropic individuals is not simply narrowed, but also sharpened (Murray et al., 2005; Murray, 2018, 2020). In states of "monotropic superdrive" (Murray et al., 2005, p. 143) finer-grain details may carry heightened relevance. It seems possible that when two monotropic individuals synchronize their "torch-beams" (Murray et al., 2005, p. 140) of intensified attention, something like a hyper-confluence of cognitive environments may occur, with increased affective reward. This may explain, for example, why in a study involving an information transfer task (Crompton et al., 2019b), autistic people both transmitted the necessary information more efficiently and experienced higher rapport when interacting with other autistic people. These findings have potential implications for how the communicative competence of autistic people is assessed, particularly if assessing interlocutors are non-autistic.

Less common, but equally as important, are the moments where the gap between sometimes very different dispositions are bridged. The familiar cross-dispositional conversations involving A1, A6, and A8, while low on flow and at times asymmetrical, demonstrate how the familiarity of an interlocutor (X1, X6, and X8, respectively) can be functionally supportive where the autistic speaker struggles. In these conversations additional processing time was given, interruptions minimized and mis-speaks accommodated for. Yet it was during the conversations with non-autistic strangers where some of the most surprising moments of connection and mutual understanding were made. A6 and A7, with their respective unfamiliar cross-dispositional interlocutors managed to reach a state of attunement, flow, and rapport through the establishing of affective common ground. In the first instance this was achieved through warm curiosity manifesting in frequent questioning about the other's experiences, and in the second through the volunteering of personal information and emotional openness.

One potential reason for the high levels of mutual understanding across all conversations may be because speakers were orientated around a central topic (loneliness) which, having agreed to participate, they had an intrinsic motivation to address. If this is the case, it is not necessarily a limitation of this study: it points to the importance of creating engaging opportunities for interaction that match an autistic person's interests in order to support communication, something that mirrors findings by Koegel et al. (2013) and Wood (2019). This is further supported by the moments in these conversations where the discovery of a shared intense by pairs of autistic interlocutors sparked significantly increased conversational flow and interpersonal attunement. Another potential reason to consider is that participants may have become more accustomed to the task across the three conversation conditions. However, for six of the eight core autistic participants (all except for A7 and A8), the matched-dispositional conversation conditions where increased flow and attunement were observed came second, not third.

Limitations and Directions for Future Research

As is often the case with rich, qualitative data, our sample size is small and would bear replication. In terms of method, the absence of a non-autistic-to-non-autistic pairing condition for the conversations may seem to be a short-coming, particularly for readers more accustomed to experimental designs. This, however, was a methodological choice. In their study analyzing patterns of intersubjectivity among small groups of autistic speakers, Heasman and Gillespie (2019, p. 910) chose to focus solely on the autistic-only interaction, arguing the following:

> Autistic people are neurologically divergent, yet methods for investigating autistic sociality tend to assume neurotypical definitions of being social. Comparative design often results in autistic behavior being interpreted as a deficit, rather than a difference, from neurotypical benchmarks (Heasman and Gillespie, 2019, p. 910).

The aim of this present study was to investigate the strength of the hypothesis that the relevance theoretic notion of mutual manifestness might serve to support the double empathy problem theory of mutual misunderstanding in cross-dispositional communication. As such, our interests centered around analyzing the way in interactions unfolded in conversation taking place between autistic and non-autistic speakers. Some form of comparison was, of course, necessary and we felt that given our interest in the role of mutual manifestness, *familiarity* served as the most meaningful condition criteria (hence core autistic speakers were asked to bring a familiar conversation partner for their first conversation, and then were paired with an autistic stranger and a non-autistic stranger). However, in future replications of this study it may be interesting to include further conversation conditions, involving pairs of familiar and unfamiliar non-autistic speakers.

Perhaps the most important limitation of this study, however, relates to the sampling of participants: of whom all were white Caucasian. This occurred organically through the self-selection of the participants, though likely also reflects both the demographic of the city within which the research took place, and the diagnostic biases against autistic people of color and minority ethnicities (Begeer et al., 2009; Fein and Rios, 2018; Jones and Mandell, 2020; Cascio et al., 2021). This matters and not only because of the urgent imperative to shift the focus of autism research away from both the Global North and white-centric stereotypes. These conversations featured a high degree of rapport, conversational flow, and mutual understanding, but this all occurred within a white, mono-cultural context. Cascio et al. (2021) have noted the "double minority status" that some autistic people of color may experience: something that may further trouble opportunities for mutual understanding by reducing what is held in common. Further studies investigating intersubjectivity or the DEP may wish to address this, and actively include autistic people of color within the cohort. Additional implications for further research include replicating this study with a larger and more diverse cohort of autistic participants, as well as exploring the longer-term impact of therapies or interventions based around shared flow states on the pragmatic and prosocial abilities of autistic individuals.

Finally, there is an important caveat to be made in relation to the present study. Findings such as these, which indicate that autistic people may enjoy more synchronous communication with fellow autistic individuals, must absolutely not be interpreted as support for the exclusion of autistic people from "mainstream" society. Furthermore, findings from this study have not suggested that cross-dispositional attunement is an impossibility: quite the opposite. We hope that these findings might contribute to efforts to support and facilitate mutually satisfying cross-dispositional interactions.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because the ethical approval given for this study by the Tier II Arts and Humanities Ethics Panel at the University of Brighton covered the publication of anonymized extracts only. This was based on the understanding that even when identifying features are redacted from transcripts, conversation in full are inherently recognizable. As such, regretfully we are unable to provide transcripts in full. Requests to access the datasets should be directed to Gemma L. Williams, glwilliamsresearch@gmail.com.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Tier II Arts and Humanities Ethics Panel at the University of Brighton. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

CRediT contributor roles: GW, TW, and CJ: conceptualization, methodology, validation, writing—review, and editing. GW: data curation, project administration, and writing—original draft. GW and TW: formal analysis and investigation. GW: funding acquisition. TW and CJ: supervision. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.616664/full#supplementary-material

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Preliminary Evaluation of the FETASS Training for Parents of Children With Autism Spectrum Disorder: A Pilot Study

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While several recent evaluation studies have shown the efficacy of parent training programs for children with neurodevelopmental disorders, manual-based training in German is still scarce. To address this gap, we developed a specific modularized training program for parents of children from preschool to pre-adolescent age with Autism Spectrum Disorder (FETASS). The overarching purpose of the FETASS intervention is to enhance social communication behavior and quality of life of the child by coaching parents. As a proximal target, the FETASS training aims to provide families with behavior management and communication strategies. The development of the training was influenced by published behavioral parent trainings and autism-specific interventions. The training comprises eight weekly sessions and targets families whose children have a diagnosis of Autism Spectrum Disorder (ASD) without intellectual and language impairments. As a preliminary pilot study, the purpose was to evaluate the acceptability of the training. Furthermore, the study aimed at initially evaluating social communication behavior, quality of life of the child, parental stress level, and parenting after training in comparison to a treatment as usual (TAU) group. Exploratively, long-term effects were investigated after 6 months of training as well. In total, 57 families participated (n[TAU] = 29, n[FETASS] = 28). Questionnaires about social communication behavior and quality of life of the child, parental stress, and parenting were administered at three time points (t1: baseline TAU/FETASS, t2: post TAU/FETASS; and t3: 6-month follow-up after FETASS). Primary outcome measures were the social communication behavior of the child and the parent's proxy report on quality of life of the child. Secondary outcome measures were changes in parental stress and parenting behavior. Acceptability of the training was very high and we had almost no dropouts during training. Results for the primary outcome measure of social communication behavior, overall quality of life of the child, and long-term effects on social communication behavior were not significant. While long-term findings for parent stress reduction and for the quality of life of the child are promising, further research has to be done in a future randomized controlled trial.

Keywords: parent training, Autism Spectrum Disorder, Freiburg Parent Training for Autism Spectrum Disorder, quality of life, preliminary evaluation, parental stress, children

INTRODUCTION

Autism Spectrum Disorder (ASD) is known as a neurodevelopmental disorder with impairments in social interaction and communication skills accompanied by restricted interests, preoccupations, or stereotyped rigid behavior. Furthermore, children with ASD represent a very heterogeneous group with a large range of functional levels and varying levels of impairment, as well as varying levels of non-impairments in the different domains of development. This fact is taken into account in the diagnostic criteria of the DSM-5 [American Psychiatric Association (APA), 2013], which makes it possible to differentiate comorbidities (e.g., with or without speech delay, with or without cognitive impairment, and with or without ADHD) and better address the individual needs of each child with ASD.

Even if a child with ASD does not have additional language or cognitive impairments, families often report difficulty with everyday social situations in areas such as social communication and interaction or because of co-occurring behaviors that challenge (Brookman-Frazee et al., 2006; Lecavalier et al., 2006). It is further known that these families report on restrictions to their quality of life (Vasilopoulou and Nisbet, 2016) and a higher level of stress (Baker-Ericzén et al., 2005; Hayes and Watson, 2013). Estes et al. (2009) emphasized that a parent's ability to manage their children's challenging behaviors is a critical target for interventions to address the child's functioning and decrease parental stress.

There is a substantial body of evidence that parental training can be effective to enhance the developmental trajectory of children with behavioral concerns (Webster-Stratton et al., 1989; Sanders et al., 2006; Weisz and Kazdin, 2010; Lee et al., 2012). As for parent-centered interventions in ASD, there has been an abundance of research on the efficacy of interventions such as Applied Behavior Analysis Approaches (for a review, see Virués-Ortega, 2010) or TEACCH (Mesibov et al., 2002; Turner-Brown et al., 2019) in which parents are involved as co-therapists. Furthermore, there are suggestions that behaviorally-oriented parent training is effective in reducing overreactivity in children with ASD (Matson et al., 2009; Whittingham et al., 2009). The efficacy of specific parent-mediated interventions for children with ASD is reviewed by Oono et al. (2013) and evidence for positive changes in patterns of parent-child interaction regarding shared attention is reported.

Rigorous randomized controlled trials (RCT) were conducted on the parent-led intervention "Preschool Autism Communication Therapy" (PACT; Green et al., 2010; Pickles et al., 2016). Herein, the parents with autistic children in the age range of 24–60 months are instructed to implement regular communication interventions at home to achieve improvements in child communicative behavior. Results of the RCT by Green et al. (2010) showed no immediate post-training effects on the ASD symptoms measured by the Calibrated Severity Score of the ADOS (CSS; Gotham et al., 2009). Yet, effects on proximal aspects of the dyadic parent-child interaction, e.g., "parental synchronous response to the child" could be found. Finally, these children showed long-term specific improvements of ASD symptoms in the follow-up evaluation 6 years after intervention (Pickles et al., 2016).

But overall, some quality concerns have been recently raised in the project Autism Intervention Meta-Analysis (AIM) about studies investigating efficacy of autism intervention in general, and behavioral intervention in particular (Sandbank et al., 2020; Crank et al., 2021).

As mentioned above, it is well-documented that parents of children with ASD show a higher level of stress (Davis and Carter, 2008; Estes et al., 2009; Hayes and Watson, 2013) and there is some evidence of a relationship between parent stress level and social affect and repetitive or restrictive behavior of the children (Harrop et al., 2016; Schutte et al., 2018). There is growing literature that dysfunctional parent-child interaction and parental stress can have a negative impact on the development of the autistic child (Crowell et al., 2019).

Accordingly, there have been efforts in international research to develop specific educational group training programs for families of children with ASD (Brereton and Tonge, 2005; Ingersoll and Dvortcsak, 2006; Chiang, 2013; Cutress and Muncer, 2013; Farmer and Reupert, 2013; Ji et al., 2014; Bearss et al., 2015; Ilg et al., 2016; Iida et al., 2018; Edwards et al., 2019). Even so, Preece and Trajkovski (2017) show that, in spite of the positive effect of parent education, only a few parental education group interventions exist.

For German-speaking countries, up to the last decade, there was a lack of manualized parent training programs for children from preschool to preadolescent age with ASD, especially for children without cognitive or speech impairment. To fill this gap, several groups developed parent training manuals. The TASK program (Fröhlich et al., 2014) addresses parents of young children from 3 to 6 years and teaches parents how to exercise communication strategies with their children. The FAUT-E (Schlitt et al., 2015) targets psychoeducation, behavioral family management, and communication strategies for parents of autistic children (from preschool-age to adolescence) with or without cognitive or speech impairment. At the same time and independently, the FETASS parent training has been established at our department of child and adolescent psychiatry. The intervention has been tailored to existing clinical process organization. It is suitable and feasible for the needs of families seeking specific intervention in our outpatient clinic and addresses children in the age range from preschool to pre-adolescence also focusing on Theory of Mind (i.e., understanding others' intentions, desires, beliefs, perceptions, and emotions, for example, in tasks of false belief or of recognizing facial expressions) and on management of critical situations (e.g., changes in setting or challenging social situations). The manualized program [FETASS: Freiburger Elterntraining für Autismus-Spektrum-Störungen (Freiburg Parent Training for ASD); Brehm et al., 2015] is based on behavioral methods that take into account parental concerns regarding the upbringing of a child with an ASD.

The overarching purpose of the FETASS intervention is to enhance the social communication behavior and quality of life of the child by coaching parents. As a proximal target, the intervention aims to improve the parent-child relationship by increasing the parents' understanding of the child as well as

to teach behavior management that takes into account the special features of the child with autism, i.e., by providing a highly organized environment. Furthermore, teaching strategies for clear family communication and e.g., exercising "Theory of Mind" abilities should enhance social skills in the child. As an important mechanism of change, we assume a family process perspective (Patterson, 1982), in the sense that an adaption of parenting is supposed to have an important impact on the child's social development and social-communicative behavior (see, e.g., Cox and Paley, 2003).

The present study is a "Phase-Two Evaluation" according to Smith et al. (2007) and aims to evaluate the acceptability of the FETASS training as a group intervention for parents of children with ASD without severe intellectual or language impairments in the age range from preschool to pre-adolescence. Furthermore, preliminary effects on social communication behavior, quality of life of the child, parental stress level, and parenting in comparison to treatment as usual (TAU) group were investigated. The hypotheses are (i) that there is a high acceptance of the training with a low dropout rate, (ii) that the training has positive effects on the social communication behavior and the quality of life of the child compared to TAU group, and (iii) that these effects persist reliably after the intervention. In this context, the TAU condition means routine clinical management in the outpatient unit (e.g., counseling, monitoring of medication and child's development). In addition to the primary outcome measures, parenting behavior and parental stress were investigated after the training.

MATERIALS AND METHODS

Participants and Intervention

The clinical study was conducted in an outpatient clinic of the Department of Child and Adolescent Psychiatry, Psychotherapy and Psychosomatics of the Medical Center of the University of Freiburg, Germany. The participants were parents/primary caretakers of a child between 4 and 15 years of age with a diagnosis of an ASD.

Our inclusion criteria were the following:

- Confirmed diagnosis of ASD (ICD-10: F84.0, F84.1, and F84.5) by an experienced clinician based on the "gold standard" instruments Autism Diagnostic Observation Schedule (ADOS-R; Lord et al., 1999; Rühl et al., 2004) and the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994; Bölte et al., 2006).
- Children without severe accompanying language impairment and without severe accompanying intellectual impairment.
- Children between preschool and pre-adolescent (mental) age.
- Full command of the German language.

Seventy-one families agreed to participate in the study. In the TAU group, one family dropped out because of a long-distance commute. Eight families in the FETASS group did not return the questionnaires after participating in the training.

In the end, the data of 57 families (29 in the TAU and 28 in the intervention group) were included in the statistical analyses (see **Figure 1**). Each family was asked to nominate a primary participating parent who would complete the training and answer the questionnaires. Sixteen families were also asked to complete the questionnaires 6 months after having finished the FETASS intervention.

As is shown in **Table 1**, most of the children of the participating parents were male and most of them met the ICD-10 diagnostic criteria for Asperger Syndrome.

All regular participants in the FETASS intervention group were mothers, six fathers out of 28 (21.4%) of the intervention group participated regularly as well.

In our sample, 48 of 57 (84.2%) had one child with ASD in the family, 9 of 57 (15.8%) had two children with ASD. None of the families had more than two children with ASD.

The FETASS program consists of eight weekly sessions. Small groups of up to eight parents are led by two therapists. Practical exercises, working in small groups, and discussion are carried out with the help of a workbook and presentation slides. During the FETASS intervention, parents work on individual goals. After every session, parents are asked to do homework. In sessions 1 and 2, the parents receive information about special features and explanatory models of ASD, especially with reference to Theory of Mind. The next step is promoting a good relationship

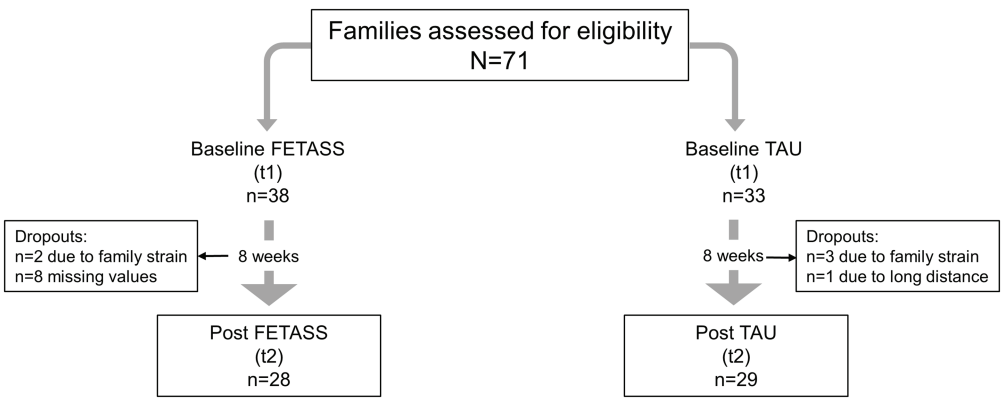

FIGURE 1 | Flow of participants.

TABLE 1 | Sample characteristics with gender and diagnoses of the children.

	TAU		FETASS	
	n	%	*n*	%
Child's gender				
Male	23	79.3	24	85.7
Female	6	20.7	4	14.3
Diagnoses				
Asperger Syndrome (ICD-10: F84.5)	14	48.3	15	53.6
Childhood Autism (ICD-10: F84.0)	8	2.6	5	17.9
Atypical Autism (ICD-10: F84.1)	7	24.2	8	28.6

TAU, treatment as usual; FETASS, Freiburg Parent Training for ASD.

with the child and perceiving the strengths of the child. The parents set individual child-centered goals they want to focus on during the training. In session 3, parents are taught to provide their child with visualizations for routines at home according to TEACCH principles (Mesibov et al., 2002; e.g., schedules, prompting strategies, visualizations, balance between demand and low arousal). Positive and negative reinforcement strategies like implementing token systems, negative consequences, or extinction are taught in sessions 4 and 5. Session 6 comprises communication strategies, i. e. promoting explicit and clear communication, prompting social situations, e.g., asking for help, or supporting the child in understanding other minds. Session 7 aims to identify and prepare critical situations. In the last session, parents are taught how to understand and manage autism-related special behavior (social interaction, restrictive interests, high repetitive activities, and sensory difficulties) and challenging situations (for an overview of the content, see also **Supplementary Material**).

Procedure

The study was approved by the Ethics Committee of the University of Freiburg (approval number: 382/14-Evaluation of the FETASS training for parents of children with Autism Spectrum Disorders) and was registered in the Deutsches Register für Klinische Studien (DRKS; DRKS-ID: DRKS00009761).

The present study was a self-financed, non-randomized clinical study with an intervention group and a TAU group. The parents either received the immediate intervention (intervention group) or TAU for 8 weeks. After the 8 weeks of TAU, the TAU group received the FETASS training for ethical reasons. The training was delivered by two therapists who were regularly supervised by one of the authors.

Parents received the questionnaires during one supplemental appointment 8 weeks before the training began. During the TAU period, the children could still be treated by a child and adolescent psychiatrist with different numbers of appointments and/or interventions (e.g., medication or other interventions). The allocation to intervention condition (FETASS vs. TAU) was not randomized, but decided according to the order of registration. We tested feasibility in terms of recruitment and retention/dropout rate.

In addition, we exploratively asked almost a third of the participants (n = 16) either of the FETASS or of the TAU group to fill out the questionnaires again 6 months after the training for an exploratory follow-up investigation (t3). There are no significant differences of the follow-up sample with respect to baseline characteristics of age, IQ, SRS-T-Total, QL-Total-LQ0-28, ESF-PS, and EFB-K-Total.

Since this was a pilot study, a sample size calculation was not performed (Eldridge et al., 2016). In order to get an accurate estimate of the SD of the outcome measure for the main trial, we followed the recommendations of Whitehead et al. (2016) who proposed sample sizes of 25 per intervention arm for small standardized effect sizes (d = 0.2) for a main trial designed with 90% power and two-sided 5% significance. Therefore, we aimed to get a pilot trial total sample size of about N = 50 participants.

Although in the literature, the effects of other parent training are frequently reported as medium or large (for ADHD: d = 0.56–0.86; see Weisz et al., 1995; Serketich and Dumas, 1996; for Triple P Stepping Stones: medium to large; see Tellegen and Sanders, 2014), we based our sample size justification on a small to medium effect, because at the beginning of our study, no effect sizes for training programs for parents with children on the Autism Spectrum have been reported.

Materials

The parents were administered the following questionnaires for evaluation:

In the Social Responsiveness Scale (SRS; Constantino and Gruber, 2005; German translation: SRS; Bölte and Poustka, 2008), the parents rate their children with respect to 65 items on a 4-point rating scale (1 = not true; 2 = sometimes true; 3 = often true; and 4 = almost always true). Scores for five scales (social awareness, social cognition, social communication, social motivation, and restrictive and repetitive behavior) and a total score (SRS-T-Total) are calculated. This questionnaire is used for a dimensional diagnostic and severity assessment of symptoms of ASD. The majority of SRS items describe social communication behavior that is associated with autistic symptoms. Psychometric properties are reported to be excellent and the measures of diagnostic accuracy as a screening instrument for ASD are very high (e.g., Fombonne et al., 2012). The retest reliability ranges from adequate to very high (according to the classification of Strauss et al., 2006). The internal consistency of the SRS-Total Scale is high; the convergent validity with well-known tests is robust.

In the Quality of Life Inventory in Children and Adolescents (ILK, Mattejat and Remschmidt, 2006), the quality of life of the child is assessed by the parent's proxy report in seven areas of daily life (with one question for each of the domains school, family, friends, alone, physical health, mental health, and overall) on a 5-point rating scale (1 = very good, 2 = rather good, 3 = partly, 4 = bad, and 5 = very bad). For these domains, the lower scores mean higher perceived quality of life of the child. Additionally, a Total Score can be calculated across all areas as LQ-Total-LQ0-28 (in this case the higher the score, the higher the reported quality of life). The retest

reliability of the Quality of Life Inventory was found to be between marginal and high, and it is suitable and often used for the evaluation of psychotherapy.

There is an ongoing debate on the different approaches for measuring changes in self-report (e.g., Meyer et al., 2013). Direct measures may have the advantage of higher sensitivity to change. Therefore, we modified the answer format of the Quality of Life Inventory in Children and Adolescents to measure change of quality of life directly. The parent's proxy report assessed whether the quality of life of their child improved or deteriorated compared to 8 weeks before on a 5-point rating scale (1 = very improved, 2 = somewhat improved, 3 = unchanged, 4 = somewhat deteriorated, 5 = very deteriorated) in the same domains as the original ILK version (Mattejat and Remschmidt, 2006). However, no psychometric characteristics are available for this new modified version.

The Parent Stress Questionnaire (ESF; Domsch and Lohaus, 2010) was developed to estimate parental life stress, role restriction, social support, and partnership. The stress level (Parental stress, ESF-PS) of the parents is assessed by 17 items asking about perceived parenting competencies (e.g., "I have doubts whether I am doing everything right in my upbringing"). Furthermore, the parents are asked in seven items about their perceived stress in the interaction with the child (e.g., "Sometimes I'm helpless about my child's behavior") and their daily parenting troubles (e.g., "I have to help my child with more daily things than I like"). The scale "role restriction" (ESF-RR) contains statements about perceived limitations associated with raising the child (e.g., "As a mother/father, I no longer have enough time for my hobbies"). The social support scale (ESF-SS) asks about support from the social environment. The internal consistency and retest reliability are adequate to very high (range of 0.76–0.92). For standardization, stanine values (1–9) were used. For parental stress and role restriction, high scores of stanine values (7–9) mean a clinically significant level. For the social support scale, low scores indicate a low level of perceived support.

The parenting questionnaire (EFB-K) is the German short-form adaptation of the Parenting Scale (PS; Arnold et al., 1993, German version by Naumann et al., 2010) that is a self-assessment scale of parenting behavior with 13 items. The endpoints describe effective or ineffective forms of certain parenting behavior in disciplinary situations, and the parents have to decide which kind of behavior they are more likely to come up with (appropriate or inappropriate parenting, e.g., "When my child behaves inappropriately, I shout at my child or I speak in a calm voice"). Each item is rated on a 7-point rating scale. A total score as well as two subscales of overreactivity and laxness can be analyzed.

Measures

Primary Outcome Measures

The SRS is an instrument that is frequently used in autism-specific evaluation studies (e.g., Reichow et al., 2013; McConachie et al., 2015; Freitag et al., 2016). Therefore, we used the SRS as the primary outcome measure to measure social communication behavior of the child. In particular, T-scores of the scales Social Awareness (SRS-T-Awr), Social Cognition (SRS-T-Cog), Social Communication (SRS-T-Com), Social Motivation (SRS-T-Mot) and Restrictive and Repetitive Behavior (SRS-T-RRB), and the Total score (SRS-T-Total) of the Scale for Social Responsiveness (SRS; Constantino and Gruber, 2005; German translation: SRS; Bölte and Poustka, 2008) were calculated.

Also, the standardized Total Score of Quality of Life (Parent report: LQ-Total-LQ0-28) was used as a primary outcome for an overall measure of Quality of Life of the children. In addition, all seven domains were used for primary outcome analyses: Quality of life in school (QL-School), in relation to friends (QL-Friends), in relation to families (QL-Family), Quality of Life in relation to interests (QL-Alone), in relation to Physical Health (QL-Physical Health), in relation to Mental Health (QL-Mental Health), and Overall Quality of Life (QL-Overall).

Secondary Outcome Measures

As a secondary outcome measure, the Quality of Life Total Score-Change (QL-Change-Total Score) was used, together with all seven scores of the domains as described above (QL-Change-School; QL-Change-Friends; QL-Change-Family; QL-Change-Alone; QL-Change-PhysHeal; QL-Change-MentHeal; and QL-Change-Overall).

The scores of parental stress (ESF-PS), role restriction (ESF-RR), and social support (ESF-SS) were used as indicators for parents' mental health and well-being.

For parenting behavior, we applied the total score of parenting (EFB-K-Total), the Overreactivity Scale (EFB-K-Overr), and the Laxness Scale (EFB-K-Lax).

Statistical Analyses

For the post-assessments, group differences as changes to baseline were analyzed by means of one-way ANOVA. Effect sizes for group differences are reported in terms of standardized mean differences (SMD): Hedges's g, rather than Cohen's d, is used as an unbiased point estimator of effect sizes (Borenstein et al., 2009) because the former enables the computation of the 95% CI, also displayed in the forest plot of the systematic review of results.

For the follow-up-assessments, ANOVAs with three repeated measurements (baseline, post, and follow-up) were conducted. In cases violating the sphericity assumption (as checked by Mauchly's test), the Greenhouse-Geisser correction was applied.

All statistical analyses were performed with SAS software, Version 9.4 (SAS Institute Inc., Cary, NC, United States). For hypothesis testing, a significance level of $\alpha = 0.05$ was adopted. Concerning missing data, complete-case analyses were conducted, i.e., no imputation methods were applied.

RESULTS

The parents' feedback at the end of the training was very positive. Altogether, 61 of 67 families (91.04%) completed the FETASS

Parent Training. The dropout rate during the parent training was descriptively lower than in the TAU group, i.e., 6 out of 67 (8.96%) vs. 4 out of 33 (12.1%). At baseline, there were no significant differences between the two groups (FETASS vs. TAU) for all primary or secondary outcome measures, for age (range: 4;9–15;0), or for intellectual abilities (range: TAU: 67–136; FETASS: 72–133; see **Table 2**).

Primary Outcome Measures

Concerning the primary outcome measures, no significant differences can be found after 8 weeks of FETASS intervention in comparison to TAU [SRS-T-Total: $F(1, 54) = 0.01$, $p = 0.940$, $g = -0.02$; QL-Total Score-LQ0-28: $F(1, 55) = 0.01$, $p = 0.912$, $g = -0.03$].

There are neither specific effects in favor of the FETASS intervention group concerning the different scales [SRS-T-Awr: $F(1, 54) = 0.21$, $p = 0.647$, $g = -0.12$; SRS-T-Cog: $F(1, 54) = 0.21$, $p = 0.647$, $g = -0.12$; SRS-T-Com: $F(1, 54) = 0.28$, $p = 0.602$, $g = 0.14$; SRS-T-Mot: $F(1, 54) = 0.09$, $p = 0.765$, $g = -0.08$; SRS-T-RRB: $F(1, 54) = 0.43$, $p = 0.514$, $g = -0.17$] nor specific short term effects concerning the domains of quality of life of the child [QL-School: $F(1, 52) = 0.05$, $p = 0.825$, $g = -0.06$; QL-Family: $F(1, 54) = 0.13$, $p = 0.721$, $g = 0.09$; QL-Friends: $F(1, 55) = 0.37$, $p = 0.544$, $g = -0.16$; QL-Alone: $F(1, 52) = 0.03$, $p = 0.855$, $g = -0.05$; QL-PhysHeal: $F(1, 55) = 0.15$, $p = 0.701$, $g = -0.10$; QL-MentHeal: $F(1, 55) = 0.14$, $p = 0.708$, $g = 0.10$; QL-Overall: $F(1, 55) = 0.09$, $p = 0.765$, $g = 0.08$].

Point estimates and confidence intervals of effect sizes for the primary outcome measures are part of the forest plot in **Figure 2**.

Secondary Outcome Measures

In the Total Score of the quality of life (QL-Change-Total Score), no significant improvements could be found [$F(1, 42) = 3.32$, $p = 0.075$, $g = 0.57$]. Quality of life in relation to "Mental Health" of the child improves significantly in the FETASS group compared to the TAU group [QL-Change-Mental Health: $F(1, 42) = 4.73$, $p = 0.035$, $g = 0.68$] after training.

Improvements in parental stress and role restriction do not reach significance [ESF-PS: $F(1, 46) = 2.39$, $p = 0.129$, $g = 0.45$; ESF-RR: $F(1, 46) = 2.29$, $p = 0.137$, $g = 0.44$]. Descriptively, the social support of the parents tends to decrease in the FETASS group [ESF-SS: $F(1, 46) = 3.70$, $p = 0.061$, $g = -0.56$].

The parenting behavior scales (Total and Overreactivity) do not achieve any significance [EFB-K-Total: $F(1, 42) = 0.69$, $p = 0.411$, $g = 0.25$; EFB-K-Overr: $F(1, 42) = 1.32$, $p = 0.257$, $g = 0.34$]. The parenting scale "laxness" shows no changes at all in both groups [EFB-K-Lax: $F(1, 42) = 0.21$, $p = 0.649$, $g = -0.14$].

For the secondary outcome measures, point estimates and confidence intervals of effect sizes are also displayed in the forest plot in **Figure 2**.

TABLE 2 | Baseline sample characteristics for quantitative variables of chronological age, intellectual abilities, and social communication behavior, quality of life, parental stress, and parenting in the TAU and FETASS group.

	TAU			FETASS			*F*	*p*
	n	*M*	*SD*	*n*	*M*	*SD*		
Age	29	10.04	2.11	28	10.52	2.53	<1	
IQ	29	98.79	15.43	28	99.43	14.04	<1	
SRS-T-Total	29	81.34	9.59	28	78.89	8.56	<1	
SRS-T-Awr	29	75.00	7.92	28	71.50	8.14	1.95	0.168
SRS-T-Cog	29	77.00	9.34	28	74.57	7.07	<1	
SRS-T-Com	29	82.79	12.68	28	80.71	11.39	<1	
SRS-T-Mot	29	75.82	9.90	28	76.46	10.17	<1	
SRS-T-RRB	29	79.90	11.47	28	78.71	10.61	<1	
QL-Total Score-LQ0-28	29	15.83	3.35	28	16.50	3.33	<1	
QL-School	28	2.93	1.36	27	2.59	0.89	<1	
QL-Family	29	2.21	0.90	27	2.41	1.01	<1	
QL-Friends	29	3.62	1.05	28	3.25	0.97	1.92	0.171
QL-Alone	29	2.48	1.30	27	2.30	1.03	<1	
QL-PhysHeal	29	2.03	0.73	28	2.11	0.92	<1	
QL-MentHeal	29	3.24	0.83	28	3.14	0.76	<1	
QL-Overall	29	2.69	0.81	28	2.68	0.61	<1	
ESF-PS	29	8.00	1.16	19	8.11	1.15	<1	
ESF-RR	29	6.90	1.59	19	7.53	1.43	1.95	0.170
ESF-SS	29	3.86	1.66	19	3.74	1.97	<1	
EFB-K-Total	29	3.01	0.82	22	3.23	1.02	1.58	0.215
EFB-K-Overr	29	3.51	1.05	22	3.97	1.26	3.37	0.074
EFB-K-Lax	22	2.57	0.90	22	2.57	1.28	<1	

TAU, treatment as usual; FETASS, Freiburg Parent Training; SRS, social responsiveness; SRS-T-Total, SRS total score; SRS-T-Awr, social awareness; SRS-T-Cog, social cognition; SRS-T-Com, social communication; SRS-T-Mot, social motivation; SRS-T-RRB, restricted interests and repetitive behavior; QL-Total-LQ0-28, quality of life total score; QL-School, quality of life in school; QL-Family, quality of life in families; QL-Friends, quality of life in relation to friends; QL-Alone, quality of life in relation to interests; QL-PhysHeal, quality of life in relation to physical health; QL-MentHeal, quality of life in relation to mental health; QL-Overall, overall quality of life; ESF-PS, parental stress; ESF-RR, role restriction; ESF-SS, social support of the parents; EFB-K-Total, parenting scale total score; EFB-K-Overr, overreactivity; EFB-K-Lax, laxness in parenting.

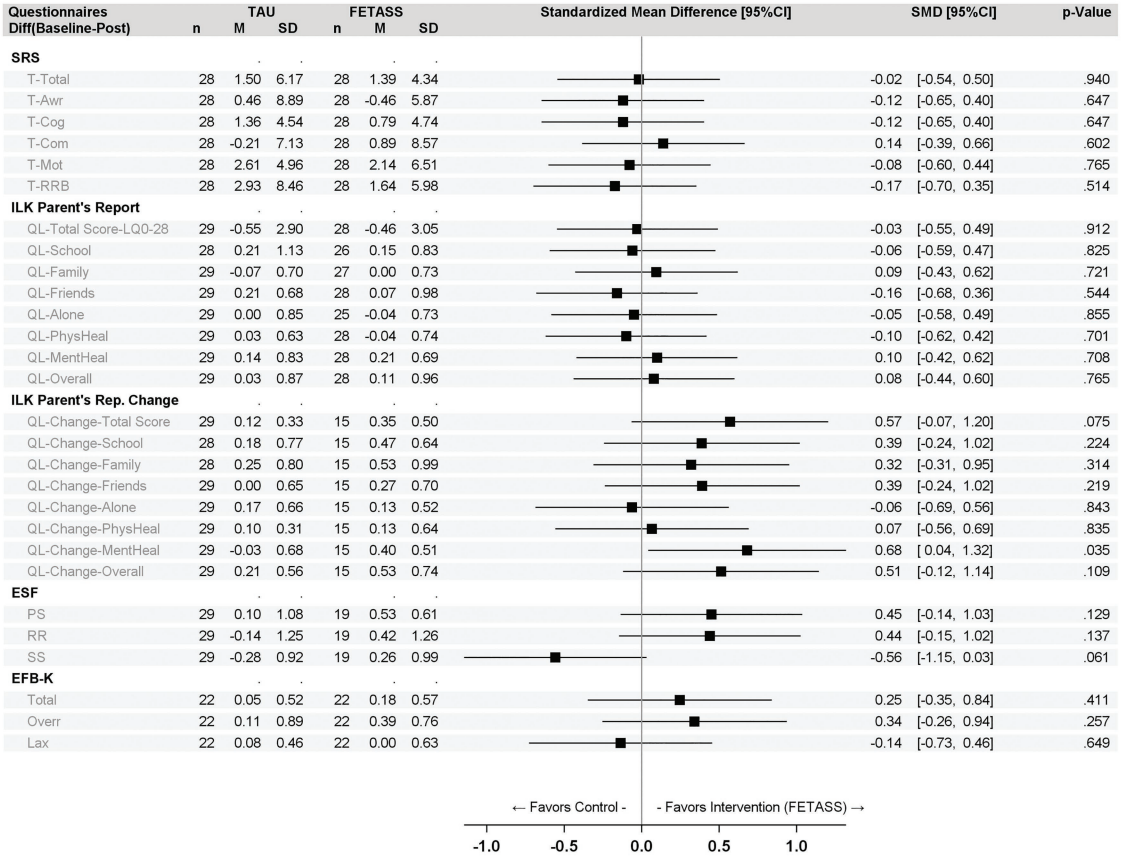

FIGURE 2 | Forest plot with point estimates and 95% CI of standardized mean differences of the primary and secondary outcome measures (for abbreviations see **Table 2**).

Long-Term Effects

For the primary outcome measures of communication behavior, a trend of improvement 6 months after training can be described, but all fall short of the adopted significance level [SRS-T-Total, $F(2, 30) = 2.61$, $p = 0.090$; see also **Table 3**]. For the follow-up measures of quality of life, there is a significant improvement [QL-Total Score-LQ0-28: $F(2, 22) = 3.81$, $p = 0.038$]. Also, the quality of life in the domain "Alone" [QL-Alone: $F(1.333, 13.332) = 4.38$, $p = 0.047$; Greenhouse-Geisser correction] shows significance, indicating an improvement in the child's ability to organize his activities by himself. Further, a significant reduction of parental stress after 6 months is obtained [ESF-PS: $F(2, 22) = 5.10$, $p = 0.015$]. The comparison between the follow-up and the two time points (t1 + t2) is significant for a reduction of parental stress over time [$F(1, 11) = 6.71$, $p = 0.025$].

The parenting measures do not show any significant changes over time [EFB-K-Total: $F(2, 22) = 0.32$, $p = 0.733$; EFB-K-Overr: $F(2, 22) = 0.80$, $p = 0.463$; PS-Lax: $F(2, 22) = 0.97$, $p = 0.395$]. Descriptively, overreactivity shows a slight trend to further decrease after 6 months (see **Table 3**).

For the direct change measurement of quality of life (see **Table 4**), significant improvements can be described in the total score and the quality of life in relation to friends after 6 months [QL-Change-Total Score: $S = 33$; $p = 0.017$; QL-Change-Friends: $S = 10.5$; $p = 0.031$].

DISCUSSION

The present pilot study aimed to investigate the acceptability of the FETASS program, a specific modularized training program for parents of children with ASD aged from preschool to pre-adolescence. Social communication behavior, quality of life of the child, parental stress level, and parenting were preliminary evaluated in a case-control comparison immediately after training and, exploratively, in a follow-up. According to Smith et al. (2007), this study can be considered as a "Phase-Two Evaluation": After manualization of the intervention, the acceptability of the manual has to be checked and a pilot case-control testing has to be conducted. In a next step, efficacy of the training must be investigated in a randomized controlled design.

Acceptability

The parents' feedback of the training intervention was positive and we had a low dropout rate during training. We interpret

TABLE 3 | Baseline (t1), post (t2), and follow-up (t3) statistics of the primary and secondary outcome measures ($N = 16$).

	Baseline (t1)		Post (t2)		Follow-up (t3)		*F*	*p*	Sign. *a priori* contrasts
	M	*SD*	*M*	*SD*	*M*	*SD*			
SRS-T-Total	79.63	12.99	79.25	11.02	76.86	11.46	2.61	0.090	
SRS-T-Awr	72.13	13.29	71.25	9.27	68.69	8.72	1.70	0.207	
SRS-T-Cog	73.88	12.45	74.31	10.69	72.06	9.90	1.71	0.206	
SRS-T-Com	79.94	12.14	80.06	11.92	78.88	14.04	0.31	0.669	
SRS-T-Mot	73.88	14.70	74.13	14.08	71.38	16.20	1.62	0.220	
SRS-T-RRB	79.88	14.33	81.19	14.02	77.63	10.56	1.62	0.220	
QL-Total Score-LQ0-28[1]	16.83	2.52	17.08	2.68	19.42	2.68	3.81	0.038*	t3 > (t1+t2)
QL-School[2]	2.80	0.79	3.00	0.82	2.60	1.07	0.92	0.399	
QL-Family[1]	2.17	0.83	2.08	0.90	2.00	0.603	0.19	0.793	
QL-Friends[1]	3.58	0.67	3.25	0.87	2.83	0.937	3.51	0.063	
QL-Alone[3]	2.36	1.21	2.45	1.03	1.72	0.90	4.38	0.047*	t3 < (t1+t2)
QL-PhysHeal[1]	2.00	0.74	2.25	0.75	1.75	0.75	1.57	0.232	
QL-MentHeal[1]	2.92	0.67	2.83	1.11	2.42	0.792	2.29	0.127	
QL-Overall[1]	2.25	0.62	2.25	0.621	2.17	0.72	0.15	0.757	
ESF-PS[1]	7.58	1.51	7.25	1.86	6.75	2.05	5.10	0.015*	t3 > (t1+t2)
ESF-RR[1]	7.25	1.29	7.83	1.40	6.75	1.96	2.88	0.088	
ESF-SS[1]	3.58	1.56	3.42	1.62	4.08	1.93	2.01	0.162	
EFB-K-Total[1]	2.90	0.82	2.77	0.77	2.78	0.81	0.32	0.733	
EFB-K-Overr[1]	3.18	1.03	3.06	0.99	2.85	0.97	0.80	0.463	
EFB-K-Lax[1]	2.72	0.89	2.65	0.76	2.88	1.09	0.97	0.395	

*SRS, social responsiveness scale; SRS-T-Total, SRS-total score; SRS-T-Awr, SRS-social awareness; SRS-T-Cog, SRS-social cognition; SRS-T-Com, SRS-social communication; SRS-T-Mot, SRS-social motivation; SRS-T-RRB, SRS-restricted interests and repetitive behavior; QL, quality of life inventory; QL-Total-LQ0-28, quality of life total score; QL-School, quality of life in school; QL-Family, quality of life in families; QL-Friends, quality of life in relation to friends; QL-Alone, quality of life in relation to interests; QL-PhysHeal, quality of life in relation to physical health; QL-MentHeal, quality of life in relation to mental health; QL-Overall, overall quality of life; ESF, parenting stress questionnaire; ESF-PS, parental stress; ESF-RR, role restriction; ESF-SS, social support of the parents; Parenting Scale, EFB-K; EFB-K-Total, parenting scale total score; EFB-K-Overr, overreactivity; EFB-K-Lax, laxness in parenting. *p < 0.05.*

[1]N = 12.

[2]N = 10.

[3]N = 11.

TABLE 4 | Quality of life (change) at the 6-month follow-up ($N = 14$).

	Follow-up (t3)			
	M	*SD*	*S*	*p*
QL-Change-Total Score	0.31	0.42	33	0.017*
QL-Change-School	0.07	1.21	0.5	1.00
QL-Change-Family	0.36	0.63	10	0.125
QL-Change-Friends	0.43	0.51	10.5	0.031*
QL-Change-Alone	0.21	0.43	3	0.250
QL-Change-PhysHeal	0.29	0.61	3	0.250
QL-Change-MentHeal	0.43	0.65	13.5	0.070
QL-Change-Overall	0.36	0.74	12.50	0.180

*Quality of Life Inventory-Change, QL-change; QL-Change-Total-Score, quality of life total score-change; QL-Change-School, quality of life in school-change; QL-Change-Family, quality of life in families-change; QL-Change-Friends, quality of life in relation to friends-change, QL-Change-Alone, quality of life in relation to interests-change; QL-Change-PhysHeal, quality of life in relation to physical health-change; QL-Change-MentHeal, quality of life in relation to mental health-change; QL-Change-Overall, overall quality of life-change. *p < 0.05.*

this as high acceptance of the program. In summary, the training appeared feasible in outpatient clinical procedures with a high acceptance from the parents. However, it should be critically noted that further important feasibility measures, e.g., qualitative assessments of outcome measures or clear criteria of acceptability according to Eldridge et al. (2016) were not collected in this study.

Social Communication Behavior

In our pilot study, no significant improvements in social communication behavior were found after the completion of the FETASS training compared to the TAU group or after 6 months in the follow-up.

Autism spectrum is a neurodevelopmental condition affecting individuals during their whole lifespan. Improvement of social communication behavior in ASD is a lengthy process that depends on a wide range of factors, such as social motivation and social cognition of the child, as well as early interventions or family factors. For these reasons, we were not surprised that changes in the social responsiveness could not be found after the short time of an 8-week parent intervention. In Pickles et al. (2016), effects on social communication behavior were found in a 6-year follow-up, but not directly after the 13 months of PACT intervention (Green et al., 2010). According to these findings and our results, we conclude that a primary outcome measure of communicative behavior after a short time of intervention is not sensitive enough. Future RCT trial should take into consideration other parameters of efficacy and long-term effects in follow-up. Especially, a dimensional measure of social responsiveness like the SRS (Constantino and Gruber, 2005) can be critically taken into consideration as the primary outcome, although the German version of the SRS (Bölte and Poustka, 2008) is widely applied and often used to evaluate social training of children with ASD (e.g., Freitag et al., 2016).

Indeed, in the autism community, there is a discussion about the appropriateness of reducing autism to a medical condition and to apply deficit-based instruments to measure the efficacy of an intervention. Proponents of the neurodiversity approach claim that interventions should not be aimed to "cure" autistic symptoms but rather to enhance interactions and communication with other people (Milton, 2014).

Autism Spectrum Disorder can be seen as a cluster of strengths and weaknesses with the characteristic of high diversity. Children with ASD show a specific way to communicate and interact with other people. The behavior problems of children with ASD regularly arise in the interaction with their environment and with neurotypical people, e.g., in families. Often, parents have problems in understanding their autistic children and in reacting appropriately. This, in turn, can be stressful for the children with ASD, and in consequence, the children show more challenging behavior, e.g., aggressive behavior, but also less social communication behavior or less social motivation with social withdrawal and more repetitive behavior.

With reference to the "SPELL-framework" of the National Autistic Society, Milton (2014) suggests that autism-specific interventions should provide important principles such as "Structure", "Positive", "Empathy", "Low-arousal", and "Links". The FETASS-program contains many aspects of this framework (e.g., teaching the parents to provide "structure" (Session 3), "positive parenting" (Session 1), or teaching parents how to provide an environment of "low arousal," e.g., by preparing critical situations (Session 6 + 7; see in **Supplementary Material**).

For a future RCT trial, it will be crucial to find appropriate measurements to assess (1) social communication behavior of the child and quality life of the child, but as well, to measure (2) positive, empathic, and structuring parenting (3) and factors of an appropriate environment.

In a future study, it could be useful to add an assessment of dyadic parent-child interaction, such as in the recent study protocol by Green et al. (2018).

In addition, recent research focuses on a new instrument to measure changes in communication behavior for autism intervention evaluation (BOSCC, Grzadzinski et al., 2016), which unfortunately was not available as we started with the project.

Quality of Life

Improvements in quality of life of the child (QL-Total) were not found immediately after training. When considering the parent's change report, a significant effect was found in the mental health of the children after training compared to TAU. Furthermore, significant long-term effects were found for the quality of life in different domains [Alone ("able to organize activities by her/himself"), Friends, and Total Score]. In conclusion, these findings are promising to intensify research about potential effects on mental health of the child after parent training.

However, since mental health and well-being is a very broad concept with multiple definitions and different measurement approaches, the current findings have to be confirmed by using other validated measurements of emotional states or behavioral problems in autism.

For a future study, the use of a direct assessment of changes or an assessment of the quality of life of the parents could also be considered.

Parental Stress

Concerning parental stress, we found no significant reduction after the training, but there was a significant reduction of parental stress level at follow-up. In gaining a better understanding of autistic behavior through training, parents seem to develop more appropriate skills to manage certain daily life situations and have a lower stress level. Even so, parents seem to need some time to implement the strategies they have learned. As a long-term effect, a lower stress level of parents might contribute to an enhancement of the child's development (Keen et al., 2010; Schutte et al., 2018; Crowell et al., 2019).

Surprisingly, parents describe a trend to decreased social support in the FETASS group just after training, which is contrary to our hypothesis. However, descriptively, this tendency is inverted in the follow-up measure showing an improvement in social support compared to baseline. A tentative explanation could be that the parents needed some time to establish more supportive conditions and to learn about the social support networks for families with children with ASD.

Parenting

No improvements in parenting like overreactivity and laxness were found.

In the Triple P evaluation for parents with ASD by Whittingham et al. (2009), effects in overreactivity and laxness of the parents right after the training were reported. However, positive effects decreased slightly over time.

In contrast, the results of the present study show no preliminary evidence toward a reduction of parents' self-reported overreactivity after the FETASS or in follow-up, which is not in line with our hypothesis. A reduced overreactivity can be considered as one aspect of positive parenting. For further research, more appropriate measurement of positive, empathic, and structuring parenting has to be found (see above).

Feedback on Training Materials and Minor Adjustments for Target Population

The materials used in the workbook appear primarily suitable for children of primary school age with no significant speech delay. Therefore, we recommend the application of the manual to parents of children with autism, who have an intellectual ability (IQ) of or above 70, without pronounced language impairment and within an age range from 5;11 to 12;11 years.

In summary, no improvements in social communication behavior or quality of life of the child after the FETASS training compared to TAU were found. However, there are some promising preliminary results for long-term follow-up, particularly regarding quality of life of the child as well as reduction of parental stress.

Limitations

The study follows the criteria of a Phase-Two Evaluation study according to Smith et al. (2007). Therefore, the most important limitation is the non-randomized design and the small sample size.

There might have been additional factors decreasing the specific effects of the training group. Concurrent factors like autism-specific therapy of the child, medication, and school assistance were not controlled for. This should be accounted for in further studies. A general problem in psychotherapy evaluation studies is that it is difficult to find an outcome measure (i) that can be easily blinded, (ii) that is not affected by subjective biases, and (iii) that has a high degree of sensitivity to change. Primary outcome measures of communication behavior and quality of life in the present study may contain categories that are too broad to detect improvements. McConachie et al. (2015) point out that suitable tools for detecting changes achieved through intervention studies of young children with ASD are scarce. Finally, the sample size has to be enlarged in order to detect both small to moderate effect sizes as well as long-term effects of the training.

CONCLUSION AND FUTURE DIRECTIONS

Although the etiology of autistic spectrum disorders is mostly attributed to genetic and neurodevelopmental factors, there seems to be growing evidence that parenting can influence aspects of the autism phenotype. In particular, positive parenting and an improved understanding of the child's needs can help parents in supporting their autistic children. (Greenberg et al., 2006; Baker et al., 2010, 2011a,b; Mandy and Lai, 2016; Crowell et al., 2019).

The important role of family characteristics is also stressed in Karst and van Hecke (2012), who propose a new transactional intervention model, which includes the influence of parents' characteristics on children with ASD. Karst and van Hecke (2012) point out that "most interventions for ASD are evaluated only in terms of child outcomes, ignoring parent, and family factors that may have an influence on both the immediate and long-term effects of therapy."

Baker et al. (2011a) mention that children with autism, like most children, are responsive to their family environment. In this line, changing the environment and family condition in providing a positive and low-arousal environment may be able to modify the communication and interaction abilities of children with ASD. There is evidence that providing a specific family environment that is suitable to the needs of the autistic child could be one important factor in contributing to a more positive social and psychological outcome (Howlin and Magiati, 2017). This study is a first attempt to address these factors.

At present, there is an ongoing ethical discussion about purposes and measurements in intervention studies (Lord et al., 2005; Smith et al., 2007; Spence and Thurm, 2010; Milton, 2014; McConachie et al., 2015). For future directions, there should be a consensus about (1) what the interventions for autism are aiming for, and (2) what kind of measurements can be used for evaluation of autism intervention. In future, other important measurements of the parent-based intervention should additionally be considered. This could be the assessment of emotional problems or stress-related reactions of the child, or measurements of family characteristics (e.g., family communication style, dyadic parent-child interaction, coping style, parental mental health problems, or life quality of the parents).

In conclusion, the present pilot study shows high acceptability of the FETASS Parent Training with a low dropout rate during the training. Although no significant changes in social communication behavior were found, the initial results are encouraging to investigate efficacy of the FETASS Parent Training in a future RCT trial. Especially, the results of our pilot study emphasize the importance of including follow-up measurements.

DATA AVAILABILITY STATEMENT

The datasets presented in this article are not readily available because of confidentiality reasons. Requests to access the datasets should be directed to bettina.brehm@uniklinik-freiburg.de.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Ethics Committee of the University of Freiburg. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

BB, MB, RR, and CF developed the study concept and designed the study with assistance of JS. BB and JS coordinated the study, recruited the participants, and completed the data collection. BB and RR planned the statistical analyses and RR carried out it with contributions of BB. All authors made contributions to the interpretation of the data. BB drafted the initial manuscript. All authors reviewed and revised the manuscript and approved the submission of the final manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.604851/full#supplementary-material

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A Randomized Case Series Approach to Testing Efficacy of Interventions for Minimally Verbal Autistic Children

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Background: Randomized Controlled Trials (RCTs) are the gold standard for assessing whether an intervention is effective; however, they require large sample sizes in order to detect small effects. For rare or complex populations, we advocate a case series approach as a more realistic and useful first step for intervention evaluation. We consider the importance of randomization to such designs, and advocate for the use of Randomization Tests and Between Case Effect Sizes to provide a robust and statistically powerful evaluation of outcomes. In this tutorial, we describe the method, procedures, and analysis code necessary to conduct robust single case series, using an empirical example with minimally verbal autistic children.

Method: We applied a pre-registered (https://osf.io/9gvbs) randomized baseline design with between-case effect size to a case series (n = 19), to test the efficacy of a novel, parent-mediated, app-based speech production intervention (BabbleBooster) for minimally verbal autistic children. Parent-rated probe scores were used to densely sample performance accuracy over time.

Results: Parents were able to reliably code their children's speech productions using BabbleBooster. A non-significant Randomization Test and small Between-Case Effect Size (d = 0.267), suggested there was no evidence that BabbleBooster improved speech production in minimally verbal autistic children, relative to baseline scores, during this brief period of intervention.

Conclusion: The current analyses exemplify a more robust approach to examining treatment effects in rare or complex populations, where RCT may be difficult or premature to implement. To facilitate adoption of this method by researchers and practitioners, we provide analysis code that can be adapted using open source R packages. Future studies could use this case series design to evaluate interventions aiming to improve speech and language outcomes for minimally verbal autistic children, and other heterogeneous and hard to reach populations.

Keywords: autism, minimally verbal, intervention, randomization, speech, parent-mediated, single case design

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INTRODUCTION

The core characteristics associated with autism are differences in social engagement and behavioral rigidity (American Psychiatric Association, 2013). Expressive and receptive language trajectories are highly heterogeneous, with an estimated 25% of autistic individuals[1] remaining minimally verbal beyond school age, indicating few or no words are spoken on a regular basis (Lord et al., 2004; Norrelgen et al., 2014). Development of speech by age five is one of the strongest predictors of functional outcome (e.g., academic qualification, paid employment, independent living, mental health) in adulthood (Szatmari et al., 2003; Howlin, 2005), yet a recent Cochrane review highlighted the paucity of robustly designed and adequately powered studies of language interventions for minimally verbal autistic participants (Brignell et al., 2018). High quality intervention studies are thus urgently required, yet the financial and logistical challenges of recruiting and testing a large sample of minimally verbal autistic participants can be prohibitive. The current study describes and illustrates the use of an alternative study design suitable for smaller heterogeneous samples: the randomized case series. We use data collected in a pilot study of a parent-mediated app-based speech production intervention, developed specifically for minimally verbal autistic children, to illustrate appropriate design and analysis techniques.

The Randomized Controlled Trial (RCT), in which a large group of participants is randomly allocated either to receive the treatment or a control condition, is considered the gold standard method with which to evaluate the efficacy of intervention trials (Sibbald and Roland, 1998; Kendall, 2003). Despite widespread adoption of RCTs with neurodevelopmental conditions, certain circumstances can make implementing an RCT difficult: the target population may be rare, difficult to recruit in sufficient numbers, and/or extremely heterogeneous (e.g., individual targets may need to vary by participant). RCTs are also costly to implement, and thus only appropriate once an advanced stage of intervention development has been reached, following the incorporation of prior rounds of piloting and feedback (Craig et al., 2006).

An additional pitfall of any between-subject design such as RCTs, is their reliance on single time-point measurements of pre- and post-intervention performance. This requires the comparison of the same outcome, measured on only two occasions. In an emerging skill, or for a population with highly variable test performance due to attentional or behavioral factors, this method risks over- or underestimating a treatment effect. The assumption that grouping participants at random will 'equal out' this measurement error may only be true in participants with a homogenous profile, which is rarely the case in neurodevelopmental conditions. Dense sampling, in which there is repeated assessment of the outcome measure both before and during the intervention, provides a more robust measurement method in populations with high heterogeneity or where individual differences are of special interest (Wilson, 2011).

A viable alternative to the RCT is the Single Case Experimental Design (Kazdin, 2019), in which each participant serves as their own control and multiple measurements are taken across at least two experimental phases, usually baseline and intervention. The overall goal is to establish a functional relationship between the intervention and a change in the dependent variable of interest. Single Case Experimental Designs come in many formats, predominantly either a phase design, where baseline and intervention measurement occasions are grouped together in sequential blocks, or an alternating design, where intervention and baseline sessions are interspersed. Features of the intervention usually guide design choice: alternating designs are best suited to interventions that work only while they are ongoing and do not have a lasting effect (e.g., tick chart for target behavior in class), whereas phase designs suit interventions where skills are built up and are expected to be retained over time.

Randomization is a cornerstone of good experimental design as it reduces extraneous confounds and increases internal validity (Barton, 2006). Single Case Experimental Designs can also incorporate randomization, for example in stimuli selection. Howard et al. (2015) advocate for the use of large stimuli sets whereby items are matched for baseline performance and randomly allocated to treatment or control conditions. The quantity of items and their randomized allocation counteracts the problem of regression to the mean, which can lead to spurious treatment effects. This is especially problematic when test performance is highly variable. This design suits word learning studies where there is a large bank of items to draw from, and works for populations that can sustain regular lengthy probes. However, minimally verbal autistic children can rarely attend for long enough to complete large sets of trials, and with speech sound learning there is only a limited number of appropriate targets to incorporate, so this approach does not suit all populations or interventions.

Single Case Experimental Designs are a widely accepted source of evidence in a number of fields such as education (Shadish et al., 2015), medicine (Vohra, 2016), and psychology (Kazdin, 2019). Despite the advantages of being low-cost, easy to implement and extremely flexible, Single Case Experimental Designs have been historically viewed as methodologically inferior (Concato et al., 2000). One reason for this is the lack of statistical tests available to evaluate their results, since they violate parametric assumptions of independence of observations and random sampling from the normal distribution. Single Case Experimental Designs were traditionally analyzed by visual inspection alone, in which observations of the outcome variable are graphed over time and aspects such as level, trend and variability are compared between experimental conditions. This approach incorporates the richness of the data whilst remaining simple and accessible (Heyvaert et al., 2015). However, the lack of objective decision-making guidelines leaves this approach vulnerable to bias and inconsistency between researchers (Matyas and Greenwood, 1990; Parsonson and Baer, 1992; Ninci et al., 2015).

There has been a renewed interest in Single Case Experimental Designs, based on numerous innovative quantitative approaches

[1] In this article, we use identity-first language (e.g., "autistic individual") rather than person-first language (e.g., "individual with autism"), as this has been highlighted as the preference of the majority autistic individuals and their families (Kenny et al., 2016).

to their analysis, which go beyond visual inspection (Manolov and Moeyaert, 2017). New methods enable researchers to use Single Case Experimental Designs to robustly test functional relationships between interventions and outcomes, and to compute effect sizes for cross-study comparison and inclusion in meta-analyses. A growing recognition of the value of Single Case Experimental Design when these analytic approaches are incorporated, has led to the establishment of new standards (Shamseer et al., 2015; Tate et al., 2016; Vohra et al., 2016). Replication of effects is crucial (Horner et al., 2005; Kratochwill et al., 2010), and can be achieved in various ways. For instance, using a single participant with three different exposures to or withdrawals of an intervention (ABAB design), or using three participants who each begin an AB phase intervention at staggered start time-points (multiple baseline design). In a multiple base line design, replication of the treatment effect across different individuals who begin the intervention at different times, is a source of internal validity.

An array of books, special journal issues, tutorials and simulations have been published in the past decade, all proffering new ways to statistically analyze Single Case Experimental Designs (see summary in Manolov and Moeyaert, 2017), with no clear consensus on a single standard approach. Furthermore, despite the heavy output of methods papers, published studies employing any of these methods are still rare. The randomization test (described below) is one innovative approach that has been employed in several Single Case Experimental Designs (Wenman et al., 2003; Schulte and Walach, 2006; Hoogeboom et al., 2012; Hwang et al., 2018; Alfonsson et al., 2019; Calet et al., 2019). In addition, the between-case standardized effect size (described below) has recently been used in meta-analysis (Barton et al., 2017). To our knowledge, a practical application that combines these methods has not yet been carried out to evaluate interventions in autistic populations.

Systematic reviews of language interventions in autism incorporating Single Case Experimental Design evidence have either been unable to generate an effect size at all (Lane et al., 2016; Mulhern et al., 2017), or have used the Percentage of Non-overlap statistic (Kane et al., 2010), which is unfortunately limited due to ceiling effects (Parker et al., 2011) and is confounded with length of baseline period (Allison and Gorman, 1993). Furthermore, Lane et al. (2016) assessed naturalistic spoken language interventions in autism for methodological quality and found that only half the Single Case Experimental Design studies (24 studies, n = 45) were of adequate quality. In summary, robust analysis measures and quality standards are still sorely lacking in the Single Case Experimental Designs describing language interventions in autism, limiting progress in research, policy, and practice.

The goal of this paper is to demonstrate a practical application of two innovative approaches to statistical analysis of Single Case Experimental Designs: (1) the randomization test, and its subsequent pooling across participants, and (2) a standardized Between-Case Effect Size (BCES), accounting for between-participant variance. These metrics are complementary to and independent of one another. We will briefly describe them, explain why they were chosen rather than potential alternatives, and address common criticisms. An in-depth mathematical and theoretical explanation of why these methods are appropriate can be found in Shadish et al. (2014a,b) and Hooton (1991).

The Randomization Test

An important way that randomization can be incorporated into Single Case Experimental Designs is by employing randomized assignment and testing functional relationships via the Randomization Test devised by Fischer (Rvachew and Matthews, 2017). This is done by randomly selecting the intervention schedule for a given Single Case Experimental Design from a pre-determined number of permissible schedules. The scope of this random assignment varies by Single Case Experimental Design type: in an alternating design, intervention allocation can be completely randomized (e.g., producing the sequence ABBABABBBBAABA, where A = baseline measurement occasion and B = intervention measurement occasion), whereas in a phase design the baseline and intervention measurement occasions must be grouped together in phases (e.g., AAAAAABBBBBBBBB). The number of permutations from which the allocated schedule is chosen will vary by design type, number of measurement occasions and any further constraints (e.g., a minimum baseline period before intervention is introduced in a phase design).

So long as the intervention schedule was randomly allocated from a number of possible permutations, a Randomization Test can be performed by computing a test statistic (e.g., the mean difference score of A versus B occasions) for each permissible permutation, via resampling. We provide an example using data from the BabbleBooster pilot project in **Figures 1**, **2** (note that raw scores are used rather than percentages). There are eight possible permutations of the intervention schedule, with a minimum of six and a maximum 13-week treatment period as illustrated in **Figure 3**). Each schedule includes 17 opportunities to assess the outcome measure; average accuracy during the baseline period (all the A weeks) is then subtracted from average performance during the treatment period (B weeks). We then generate the range of all eight possible mean difference scores (assuming the intervention had started at session 5, 6, 7, 8, 9, 10, 11, or 12) and compare them in size to the actual mean difference obtained. If the intervention had no effect (the null hypothesis), there would be a 1/8 chance that the obtained mean difference would be the greatest score when compared to each and all of the seven other outcomes. The relative ranking of the actual mean difference is thus translated into a p-value, for example, if there are eight possible comparisons, and there are five hypothetical outcomes with the same or greater mean difference, this equates to a p-value of 5/8 or 0.625.

Conceptually, random assignment strengthens internal validity by counteracting the threats of maturation and history (Heyvaert et al., 2015). The Randomization Test is not linked to a specific test statistic, so if the mean difference is not appropriate, there is flexibility to use a different metric. As a non-parametric test, the Randomization Test is robust to violations of certain assumptions that are difficult to meet in Single Case Experimental Design research, namely independence

1. Select a random allocation schedule from all permissible options – in this case Participant 1 receives Permutation 4

Measurement occasions

Permutations	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17
1	A	A	A	A	B	B	B	B	B	B	B	B	B	B	B	B	B
2	A	A	A	A	A	B	B	B	B	B	B	B	B	B	B	B	B
3	A	A	A	A	A	A	B	B	B	B	B	B	B	B	B	B	B
4	A	A	A	A	A	A	A	B	B	B	B	B	B	B	B	B	B
5	A	A	A	A	A	A	A	A	B	B	B	B	B	B	B	B	B
6	A	A	A	A	A	A	A	A	A	B	B	B	B	B	B	B	B
7	A	A	A	A	A	A	A	A	A	A	B	B	B	B	B	B	B
8	A	A	A	A	A	A	A	A	A	A	A	B	B	B	B	B	B

2. Run experiment using this schedule and measure the outcome variable (score out of 9)

1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	3

3. Compute test statistic (mean difference B-A)

A	B	
2.2	4.7	= 2.5

4. Compute mean difference for all potential permutations

Measurement occasions

Permutations	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	A	B	B-A
1	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.3	4.1	1.9
2	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.3	4.1	1.9
3	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.2	4.4	2.2
4	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.2	4.7	2.5
5	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.2	4.7	2.5
6	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.2	4.7	2.5
7	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.3	5.0	2.7
8	4	1	2	4	2	-	2	2	-	-	3	4	6	5	-	6	6	2.5	5.2	2.7

5. Count number of permutations with outcomes >= actual test statistic and divide this by total possible permutations

$$\frac{5}{8} \quad \text{p=.625 (one-tailed)}$$

FIGURE 1 | Steps needed to calculate a Randomization Test. **(1)** Random selection of intervention schedule; **(2)** repeated measurement of outcome variable; **(3)** calculation of mean difference between intervention and baseline scores; **(4)** compute all potential mean differences (one for each permissible intervention schedule); **(5)** compare the actual mean difference with all possible outcomes to obtain a rank, e.g., the fifth greatest mean difference out of eight possibilities, which corresponds with a *p*-value of 5/8 or 0.625.

of observations and random sampling from a normal distribution (Hooton, 1991). Single Case Experimental Design observations usually have a degree of serial dependency, or autocorrelation, and can display trends (Solomon, 2014); the Randomization Test can accommodate linear trend better than a group design (Michiels and Onghena, 2019).

Despite these advantages, randomization remains rare in Single Case Experimental Designs (Heyvaert et al., 2015).

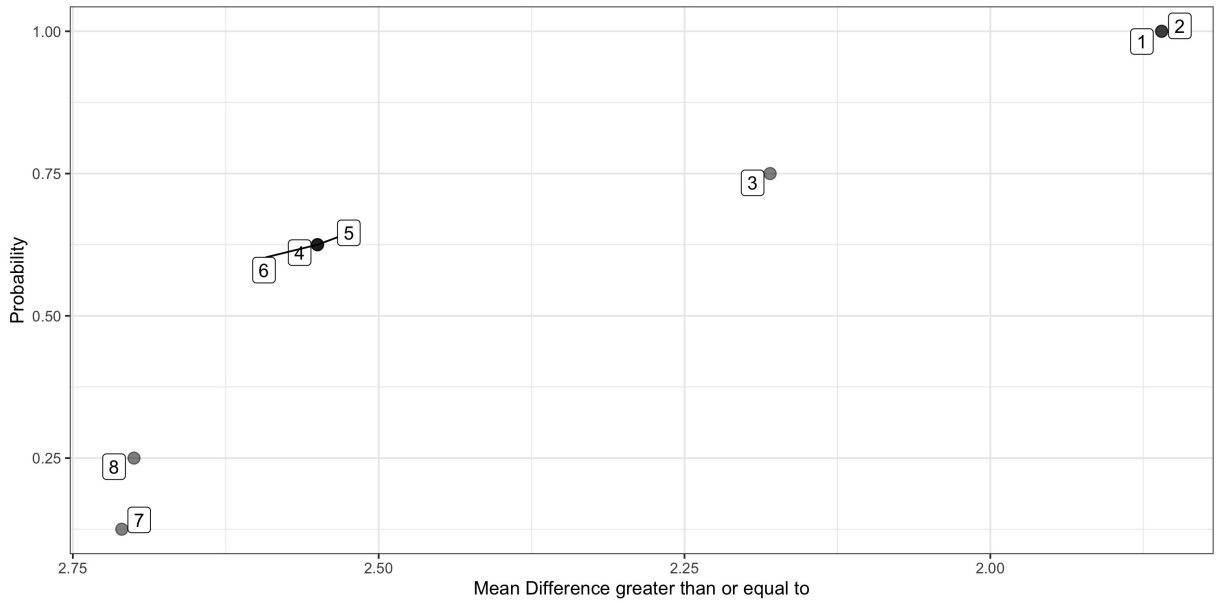

FIGURE 2 | Probability distribution of all possible mean differences. Plots the mean difference for each of eight permissible permutations in rank order, against the likelihood of the mean difference being at least as great, e.g., all mean differences are greater than 1.86, $p = 1$ that any of the eight selected at random will be at least 1.86. Only 1 is greater than or equal to 2.71, therefore the associated if the actual observed mean difference was 2.71 is $p = 1/8$ or 0.125. Data points are labeled according to the permutation number.

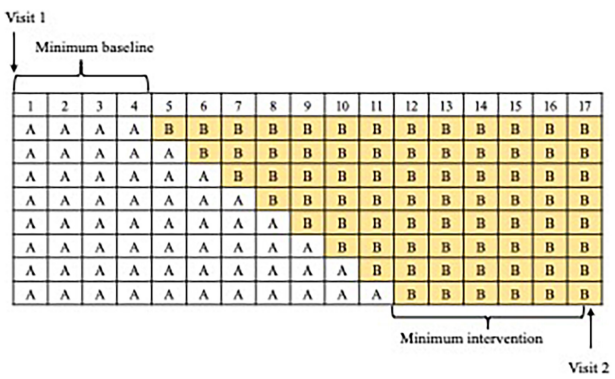

FIGURE 3 | All possible permutations of baseline (A) and intervention (B) weeks.

One criticism is that the Randomization Test's power to detect an effect diminishes in the presence of certain non-linear trends such as a delayed intervention effect, a learning curve or an extinction burst (Sierra et al., 2005; Wilson, 2011; Levin et al., 2017). Another issue is that random-assignment of intervention start point is not always possible or desirable. The pre-determined introduction point of an intervention is at odds with response-guided experimentation (Kazdin, 1980), and can be challenging if it is not known how long a stable baseline will take to achieve. Rvachew and Matthews (2017) also highlight the ethical dilemma of potentially giving some participants a very long baseline with many repeated measurement obligations prior to receiving the intervention. However, each participant does receive some exposure to both conditions, unlike an RCT where participants may be assigned to the control group and not receive any of the intervention.

As is evident from the example in **Figure 1**, if there are only eight possible permutations for a given participant, the lowest achievable *p*-value for a Single Case Experimental Design is 0.125, or 1/8, assuming a one-tailed analysis. A single AB phase Single Case Experimental Design alone is unlikely to have adequate power to detect small improvements in the target measure (Haardörfer and Gagné, 2010; Michiels and Onghena, 2019). Ways to increase power include increasing the number of measurement occasions, or replicating the result by pooling results across participants. *P*-values derived from individual Randomization Tests can be pooled across participants in a case series or multiple baseline design, to determine the likelihood of these p-values occurring by chance, using Stouffer's Z statistic (Rvachew and Matthews, 2017).

The Between-Case Effect Size (BCES)

The Randomization Test assesses the significance of a functional relationship between the intervention and a change in the outcome variable, but does not inform us as to the magnitude or variability of this effect. Effect sizes not only convey this important information, but due to their standardization, enable the comparison of effects across studies. Effect sizes are increasingly considered to be more important than *p*-values

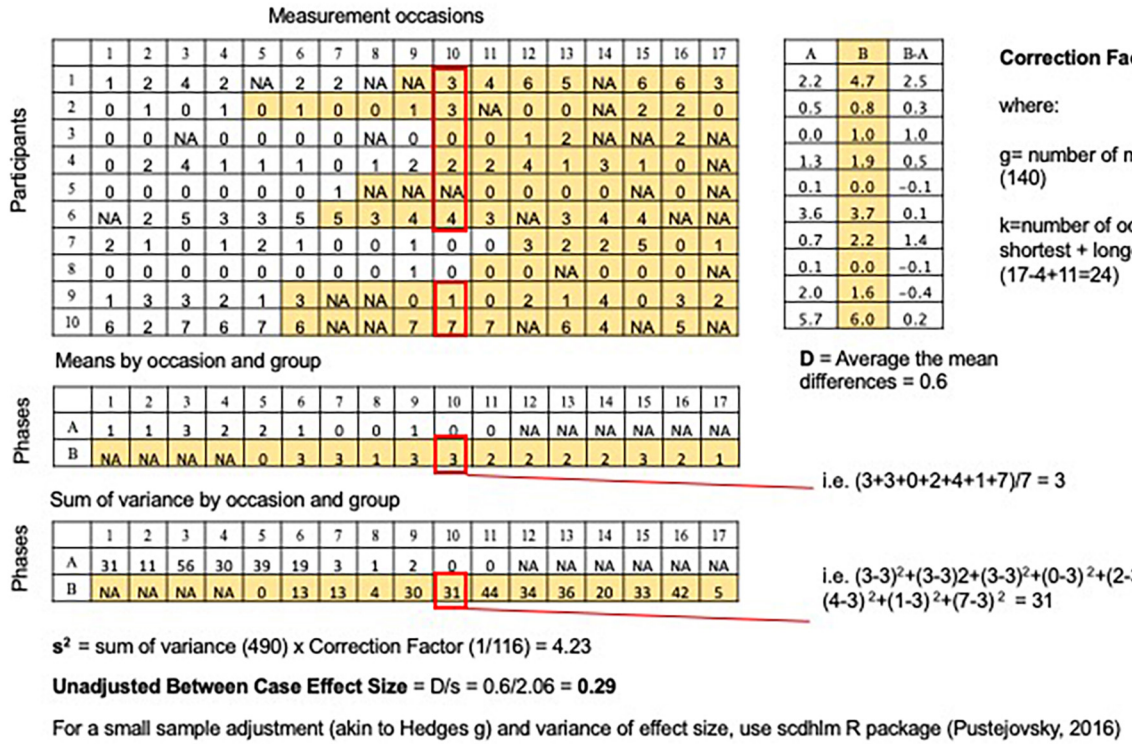

FIGURE 4 | Calculation of unadjusted Between-Case Effect Size. For each measurement occasion, group scores into occasion type (baseline or intervention) and calculate variance; sum all the variances and multiply by a correction factor; take the square root to calculate the denominator (s); numerator is the average mean difference across participants (D); effect size = D/s.

for interpreting intervention results and informing evidenced-based practice (Wilkinson and Task Force on Statistical Inference, 1999). RCTs have an established standardized effect size, Cohen's *d* (Cohen, 1977), which can be adjusted to Hedges *g* (Hedges, 1981) for small samples. The unit of comparison is standard deviations of outcome variable. Effect sizes historically developed for Single Case Experimental Designs cannot be standardized in the same way and do not account for between participant variance, in the way that Cohen's *d* does in a group study (see Odom et al., 2018 for a summary of previous approaches and their limitations). The importance of determining a robust effect size for Single Case Experimental Designs is increasingly recognized (Shadish et al., 2014a), as few studies currently report effect sizes or their variances (Jamshidi et al., 2018).

Many effect size metrics have been proposed for single case experiments (Manolov and Moeyaert, 2017), yet there is no consensus on the best approach. Approaches using regression coefficients as effect sizes have been devised (Moeyaert et al., 2014; Shadish et al., 2014c). These are able to account for linear or non-linear trends in the data as well as for dependent error structures, however, they are more complicated to implement and interpret, when compared to mean difference based approaches (Heyvaert et al., 2015). Other approaches have been developed and tested using a Bayesian framework (Jones, 2003; Swaminathan et al., 2014; de Vries et al., 2015; Odom et al., 2018), however, implementation is similarly complex. Non-parametric approaches have been proposed such as the Randomization Test Inversion, which exploits the equivalence between a hypothesis test and a Confidence Interval to create an effect size based on the Randomization Test (Michiels et al., 2017), but this is yet to be robustly tested. Tau-U, based on the tradition of examining non-overlap between experimental conditions, combines existing non-parametric tests Mann–Whitney U and the Kendall Rank Correlation coefficient (Parker et al., 2011).

In the current study we focus on the Between-Case Effect Size (BCES) devised by Hedges et al. (2012, 2013) and Pustejovsky et al. (2014), illustrated in **Figure 4**. The BCES is easy to interpret, has been tested in simulations (Hedges et al., 2012), meta-analyses (Barton et al., 2017), tests of practical applicability (Odom et al., 2018), and comparisons with other approaches (Shadish et al., 2016; Odom et al., 2018). It is accessible to non-statisticians, given the straightforward conceptualization (based on Cohen's *d*) and the availability of several R packages (Bulté and Onghena, 2009, 2019; Pustejovsky, 2016) and primers (Hedges et al., 2012, 2013; Valentine et al., 2016) to aid calculation.

We applied this approach to evaluate a parent-mediated app-based speech production intervention for minimally verbal autistic preschoolers (n = 19). We have recently described the methods, analysis, and challenges to implementing this approach in a population of children that is difficult to recruit and has highly variable patterns of language growth (Saul and Norbury, 2020b). To our knowledge, random assignment and

between-case effect size analysis have not previously been applied to a Single Case Experimental Design targeting expressive language growth in minimally verbal autistic children. Single phase was considered the most appropriate format (rather than phase reversal or alternating), since the aim of the intervention is to teach speech sound skills, which once acquired should remain part of the child's speech sound repertoire. Employing an app-based intervention facilitated remote, repeated sampling of the outcome measure, which is a core component of Single Case Experimental Design. Indeed, the practicality of repeated sampling, and the ability to introduce blinding or independent validation into this process is a key challenge in Single Case Experimental Designs (Smith et al., 2007), which can be addressed using apps in everyday settings.

The overarching goal of the current study is to illustrate how Single Case Experimental Designs with random-assignment can be used to evaluate interventions, particularly for minimally verbal autistic children, by employing the Randomization Test and the Between Case Effect Size. To do this we use real data gathered as part of the BabbleBooster pilot project, with shared data and code (Saul and Norbury, 2020b). We illustrate how in this intervention parents could gather reliable speech attempt data, facilitating remote dense sampling using the app. All objectives and hypotheses relating to the BabbleBooster pilot project were pre-registered[2,3].

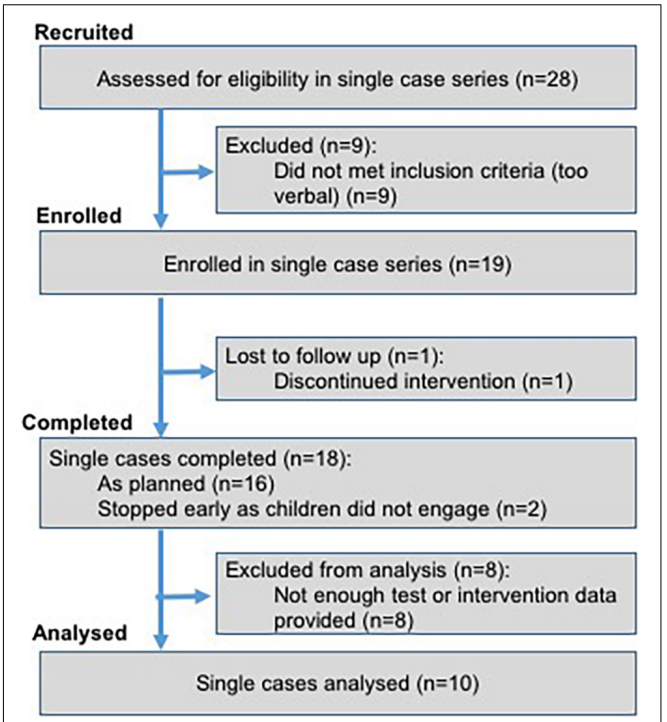

FIGURE 5 | Recruitment flow chart.

MATERIALS AND METHODS

Study Design

The study utilized an AB phase design with randomized baseline allocation; the number of weeks of baseline testing (A weeks) and the number of weeks of subsequent intervention (B weeks), were determined randomly for each participant.

Constraints on randomization were as follows:

- each participant had a minimum of three baseline (A) weeks
- each participant had a minimum of six intervention (B) weeks

These constraints were determined due to the limited timeframe available for the intervention (16 weeks), and prioritizing intervention weeks whilst retaining a long enough minimum amount of A weeks for a baseline to be established (Horner et al., 2005). Taking account of these constraints yielded eight possible intervention schedules (**Figure 3**); a different schedule was randomly assigned to each participant.

Intervention

The BabbleBooster intervention app was designed to deliver predictable and repetitive speech models via video-modeling and cued articulation (Saul and Norbury, 2020b). The app-play is parent-mediated, so parents are required to watch the stimuli with their children, encourage them to make the sound, and then provide feedback on the accuracy of the production attempt in order to trigger the reward videos. Reward videos were designed with a gradient response, so a 'good try' at a sound (an incorrect attempt) will result in a lesser reward than an accurate response. The families were encouraged to make or upload their own reward videos, based on their understanding of the individual child's interests and reward. Acceptability data and development of the app prototype are discussed in Saul and Norbury (2020b).

Participants

Figure 5 describes the process through which participants were selected for the study. Participants were 19 minimally verbal autistic children (three girls, 16 boys) for whom parents reported fewer than 10 sounds or 20 words or produced fewer than five spontaneous words during an initial assessment visit. We gathered quarterly reports on the type and amount of therapy received by each participant. Participants received an average of 0.68 h of Speech and Language Therapy per week (range: 0–2.5 h).

The children were aged 47–74 months at Visit 1 (mean = 60, *SD* = 7) with a confirmed diagnosis of autism. The following exclusions applied at initial screening: epilepsy; known neurological, genetic, visual or hearing problems; English as an Additional Language. Participants were recruited via social media, local charities, independent therapists and a university-run autism participant recruitment agency, and all took part in a larger longitudinal study (Saul and Norbury, 2020a). Ethical

[2]https://osf.io/9gvbs

[3]In light of non-significant main findings, the final section of pre-registered analyses was not carried out, as these sought to identify potential moderators of success.

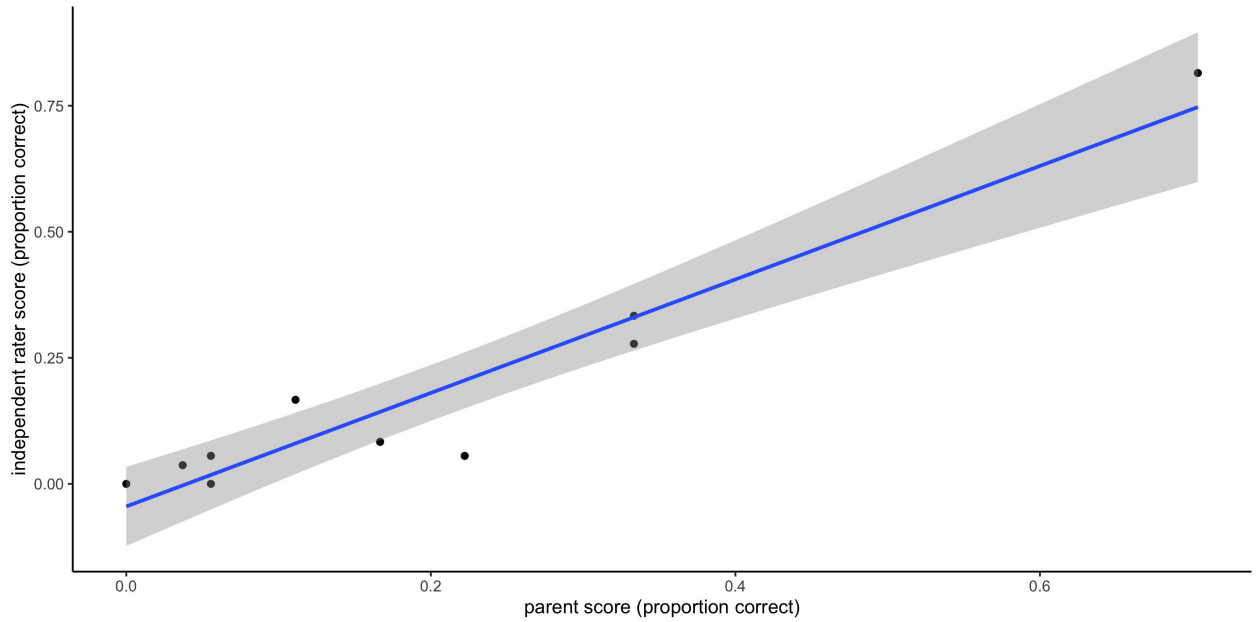

FIGURE 6 | Reliability of parent-rated versus clinician-rated weekly scores.

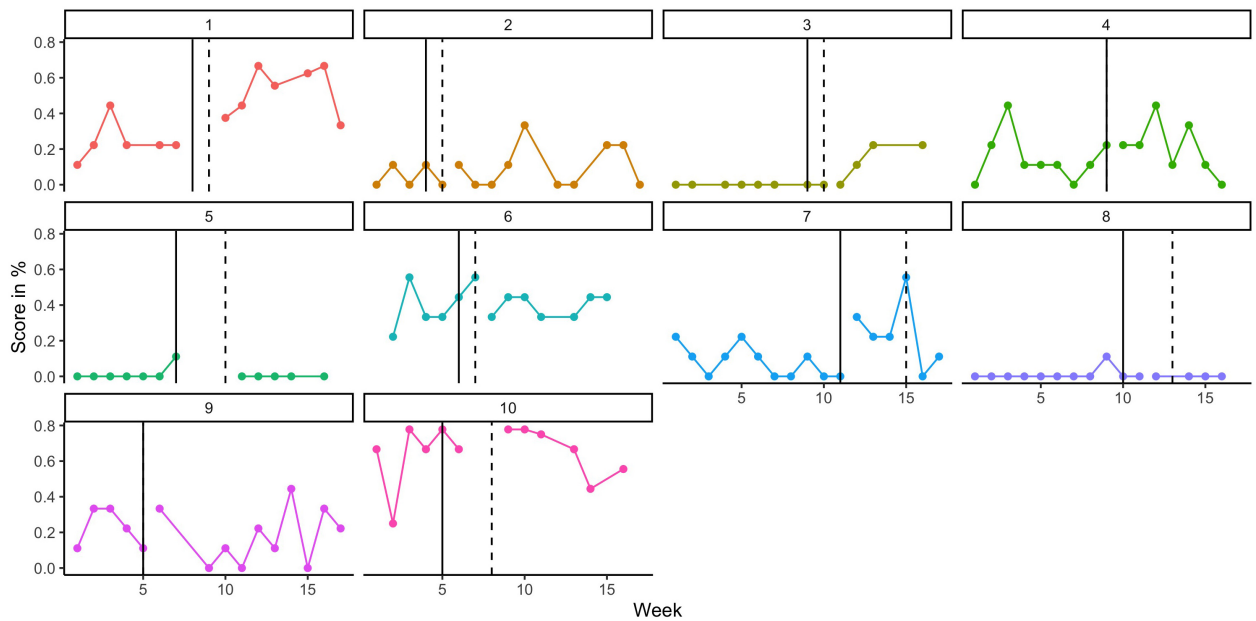

FIGURE 7 | Weekly scores on elicited phoneme test, by participant (as %). The vertical line represents the allocated start week for intervention and the dashed line is the actual start week.

approval was obtained from the UCL Research Ethics Committee (Project ID 9733/001) and informed consent was sought from parents on behalf of each participant.

Parents reported 17 participants to be White, one to be Asian and one to be Mixed Race. Eight caregivers had completed high school, eight completed university education and three completed post-graduate studies or equivalent. Eighty-eight percent of parents reported that their child had an Education Health and Care Plan, a legal document that specifies special educational support required for the child, at Visit 1.

Power

Given the above described constraints (16 weeks of data collection, 8 potential intervention schedules and 19 eligible

TABLE 1 | BabbleBooster parent rating buttons.

Button	Meaning	Example	Consequence
Yes	Child has produced elicited sound accurately	Child is asked to say /b/ and they say /b/	'Well done' video
Good Try	Child tried to make a sound but did not make the target sound	child is asked to say /b/ and they say /w/	'Good try' video
Try Again	Child does not attempt to make any sound	child is silent/shouts/cries	No video clip

TABLE 2 | Descriptive variables.

Measure	Description	Time	*n*	Mean	*SD*	Min	Max
Age	Age in months	Visit 1	18	61.6	7.5	47.6	74.6
		Visit 2	18	65.7	7.3	52.2	78.3
Receptive language	Oxford CDI words understood (Hamilton et al., 2000) (words)	Visit 1	18	182.2	135.2	5.0	406.0
		Visit 2	18	195.0	141.9	5.0	417.0
Expressive language	Oxford CDI words spoken (Hamilton et al., 2000) (words)	Visit 1	18	4.5	6.4	0.0	19.0
		Visit 2	18	11.6	26.3	0.0	90.0
Consonant inventory	CSBS Scale 11 (Wetherby and Prizant, 2002) (raw score)	Visit 1	18	6.4	3.6	1.0	13.0
		Visit 2	17	5.2	4.4	0.0	16.0
Autism symptom severity	CARS (Schopler et al., 1988) raw score	Time 1	19	42.7	4.9	35.0	52.5
NVIQ	Visual Reception and Fine Motor subtests of Mullen Scales of Early Learning (Mullen, 1995) transformed into Developmental Quotient (developmental age in months/age in months)	Time 2	19	0.36	0.13	0.13	0.56

CARS, Childhood Autism Rating Scales; CDI, Communicative Development Inventory; CSBS, Communication and Symbolic Behavior Scales; ESCS, Early Social Communication Scales; NVIQ, non-verbal intelligence quotient; SD, standard deviation; Time 1: 12 months prior to Visit 1; Time 2: 8 months prior to Visit 1.

participants) a sensitivity power analysis was conducted using simulation. One important unknown variable was how correlated dependent variable scores would be within participant, so three scenarios were modeled: low correlation (ICC = 0.25), medium (ICC = 0.50), and high (ICC = 0.75). This suggested adequate power to detect effect sizes of 0.48 and above (high correlation) to 0.84 and above (low correlation), whereas group studies of a comparable size would require larger effect sizes to reach the same power (see **Supplementary Appendix B**).

Procedure

Children were seen in their homes for two sessions (Visit 1 and Visit 2), separated by 4 months each (mean = 4.0, *SD* = 0.3). A thank you gift of a small toy or £5 voucher was provided following each visit.

At **Visit 1**, each participant received a new Samsung Galaxy Tab A6 tablet containing the BabbleBooster app[4], unless parents expressed a preference to use the app on their own Android device (n = 3). Parents were given a demonstration of the app by the experimenter, and an information pack explaining how to download and use the app. Secondly, the Probe Phonemes were selected by following the 'Sound Target Protocol' (see **Supplementary Appendix A**) and each parent-child dyad was informed of their randomly allocated intervention start date. Probe Phonemes constituted the outcome variable and comprised nine speech sounds that were elicited each week in the baseline and intervention periods. They also formed the list from which an initial three target phonemes were drawn for the intervention. Probe Phonemes remained the same for each participant and were not manipulated as part of the experiment, rather they were a necessary feature to accommodate the fact that each participant had a unique profile of speech related difficulties.

Between Visits 1 and 2, text message reminders were sent to parents to remind them of the weekly probe day, and if necessary, missed probes were rearranged for the following day. Parents also received a reminder text on the intervention start date. Thereafter, parents were asked to engage their child in play with the app for 5–10 min per day, 5 days per week. This resulted in children carrying out the intervention for between 6 and 13 weeks (see **Figure 3**). For each weekly assessment of the outcome measure, all pertinent information was uploaded to the server [date stamp, phoneme, attempt number, parent rating (either "correct," "incorrect attempt," or "no attempt")] and a video clip of the attempt. Parents pressed one of three buttons to assign a rating to the attempt, in accordance with **Table 1**.

On Visit 1 and 2, additional parent-report language measures were obtained to characterize the number of words understood and spoken by the child, as well as direct recording of the number of consonants uttered by the child during a natural language sample (Consonant Inventory).

Data collected prior to Visit 1: As the participants were drawn from a previous longitudinal study (Saul and Norbury, 2020a), further background measures, which were gathered between 8 and 12 months prior to the current study, were also available to characterize the sample. **Table 2** displays descriptive variables for the intervention group.

[4]One participant received a comparable second hand Nexus 7 tablet.

TABLE 3 | Comparison of A and B week elicited phoneme scores.

ID	A week mean (*SD*) elicited phonemes (proportion correct)	B week mean (*SD*) elicited phonemes (proportion correct)	Mean difference (B – A weeks)	Rank	*p*-value
1	0.241 (0.109)	0.525 (0.140)	0.284	3	0.375
2	0.044 (0.061)	0.110 (0.122)	0.065	4	0.500
3	0.000 (0.000)	0.139 (0.106)	0.139	2	0.250
4	0.148 (0.136)	0.206 (0.149)	0.058	2	0.250
5	0.016 (0.042)	0.000 (0.000)	–0.016	5	0.625
6	0.407 (0.135)	0.397 (0.059)	–0.011	7	0.875
7	0.148 (0.155)	0.056 (0.079)	–0.093	1	0.125
8	0.009 (0.032)	0.000 (0.000)	–0.009	6	0.750
9	0.222 (0.111)	0.178 (0.159)	–0.044	4	0.500
10	0.642 (0.196)	0.660 (0.137)	0.019	3	0.375

Primary Outcome Measure: Elicited Phoneme Weekly Score

Each child received a probe score out of 9 for each of the 16 weeks between Visit 1 and Visit 2. This was used to generate a mean baseline probe score and a mean intervention probe score, as well as the mean difference between these two measures.

Missing Data

In the pre-registered analysis, we planned to impute all missing data for the outcome variable following Enders (2010); however, following data collection we made a distinction between participants who did not reliably engage with the testing regime ('low users') and those who did ('high-users,' who each provided more than 66% of all data points). Results were reported for high-users only, both on the basis of the incomplete dataset and pooled estimates from 40 multiply imputed datasets, created using the Amelia package in R (Honaker et al., 2011). Given that using multiple imputation programs may not be feasible for all clinicians or researchers seeking to use these methods, we provide code with and without imputation in **Supplementary Appendix C**.

Reliability of Parent Ratings

The primary outcome measure is derived from parent ratings of elicited phoneme attempts. To assess reliability of parent scores, 20% of the probes were coded by a qualified Speech and Language Therapist, who was not involved in the study, and was blind to the intervention targets and individual assessment point.

To calculate the reliability of the parent ratings, we derived a list of the filenames of all available video clips downloaded from the BabbleBooster server for the 10 analyzed participants (n = 1,120). This number did not correspond with the total number of parent ratings (n = 1,248) due to the loss of some videos due to technical problems with the devices used. For coding purposes, data from incomplete weeks were also removed (n = 113). Videos were not selected completely at random: the sample needed to include at least 2 complete weeks of data for each user (n = 214 videos) since the variable we were comparing across raters was the weekly score. Weeks were chosen at random from the available weeks and comprised at least one A and one B week[5]. For each video clip, the blind coder was told which sound the child was attempting and told to rate it as 'no attempt,' 'incorrect attempt,' or 'correct attempt' in accordance with **Table 1**, corresponding to a score of 0, 0.5, or 1.

This process generated two to three randomly selected weekly scores for each of the 10 'high use' participants, which were used to compute an intra-class correlation coefficient, using the intra-class correlation ICC() command in the psych R package (Revelle, 2018). An agreement of 0.85 or higher was considered an acceptable level of agreement (Koo and Li, 2016, suggest > 0.75 represents good agreement).

Attrition and Adherence

We report adherence to allocated intervention start date for each participant, given its importance to the accuracy of the randomization test. In addition, participants were required to submit > 66% of weekly test data-points to be included in the analysis of primary outcome; proportion of missing data is reported below.

Analysis Plan

Randomization Test

The statistical model used to analyze the significance of a positive change in the primary outcome variable (elicited phoneme test score), was the randomized phase design with resampling as outlined in Rvachew and Matthews (2017). This is a one-tailed analysis, and was calculated in R (R Core Team, 2017) using the script detailed in **Supplementary Appendix C**. The anonymized dataset is available to download here: https://osf.io/rzuwt/.

P-values were pooled across participants, to gauge the consistency of any treatment effects. This was done using the sumz function in the MetaP Package in R (Dewey, 2019), which uses Stouffer's z-trend procedure to generate a *p*-value that denotes the likelihood of achieving a series of *p*-values merely by chance. We used a *p*-value of less than 0.05 for significance testing for the meta-analysis of *p*-values.

[5]Not possible for one participant due to technical problems with uploading in initial weeks.

TABLE 4 | Individual characteristics of 'high users.'

ID	Mean SLT hours/week	Autism Severity (CARS, Time 1)	NIVQ (Time 2)	Age at Visit 1	RCDI at Visit 1	ECDI at Visit 1	Consonant inventory at Visit 1	Age at Visit 2	RCDI at Visit 2	ECDI at Visit 2	Consonant inventory at Visit 2
1	1.00	35	0.38	74.6	68	0	5	78.3	51	0	1
2	1.75	41.5	0.4	61.2	290	0	4	65.1	304	1	7
3	0.66	49	0.49	56.6	5	0	5	61.4	5	0	10
4	0.50	46	0.48	60.3	282	1	7	64.0	277	1	6
5	0.02	48.5	0.28	57.2	38	0	2	60.8	47	0	5
6	0.98	37	0.56	54.4	212	19	4	58.6	224	3	4
7	0.01	43	0.38	69.6	337	0	9	73.3	412	0	7
8	0.38	46.5	0.17	62.6	8	5	12	67.0	11	1	0
9	1.25	46.5	0.13	59.5	55	0	4	63.8	65	0	1
10	0.75	37	0.53	59.8	314	9	6	63.4	327	90	16

CARS, Childhood Autism Rating Scales; ECDI, Expressive Communicative Development Inventory; NVIQ, non-verbal intelligence quotient; RCDI: Receptive Communicative Development Inventory; SLT, Speech and Language Therapy; Time 1: 12 months prior to Visit 1; Time 2: 8 months prior to Visit 1.

Between-Case Effect Size

Between-case Effect Size was calculated for the case series using the 'scdhlm' package (Pustejovsky, 2016) and following the guidelines set out in Valentine et al. (2016). Thus performing the command MB_effect_size() generated an adjusted *d* statistic as well as its variance. Sample code is provided in **Supplementary Appendix C**.

RESULTS

Reliability of Parent Ratings of Speech Production Attempts

The intra-class correlation coefficient for speech production ratings by parents compared with those by an independent rater was 0.84 when scores of 0, 0.5, and 1 were considered (0 = no response, 0.5 = incorrect attempt, and 1 = correct). When scores were re-categorized to reflect a binary correct/incorrect split (scores of 1 and 0 respectively, with an incorrect attempt scoring 0 instead of 0.5), this figure rose to 0.95. In light of this, scores of 0 and 1 were used in all subsequent analyses, rather than 0, 0.5, and 1, as originally planned. Individual weekly scores from the reliability analysis are plotted in **Figure 6** to demonstrate the level of consistency achieved. The within-participant variability of scores was also of interest, given the importance of stability in the dependent variable to the statistical power suggested in **Supplementary Appendix B**. One advantage of dense sampling is that it increases power, particularly when each participant's dependent scores are highly stable. In the current study, each participant supplied at least 12 weeks of probe data; the intra-class correlation coefficient for these scores was 0.75, signifying high consistency in production from week to week.

Randomization Test

Attrition for the randomization test was 47%, as of the 19 original participants, only 10 were classified as 'high' users of the app, insofar as they completed > 66% of test trials. Amongst these high users, the mean number of test trials completed was 82% (*SD* = 11%, range = 69–100%). It was possible to calculate efficacy measures using the data collected from these 10 participants despite the missing data points. Comparison of allocated intervention start date and actual intervention start date revealed a mean delay of 1.4 weeks (*SD* = 1.3, range = 0–3).

Figure 7 presents the individual weekly probe scores of each participant (score out of 9 expressed as a percentage). These scores were used to compute the mean difference score for each participant and compare it to the distribution of potential outcomes. Intervention was deemed to commence at the actual rather than allocated start date. **Table 3** reports each participant's mean score and standard deviation for A and B weeks, the mean difference between them, and the corresponding rank and *p*-value associated with that mean difference. A non-significant Stouffer's *Z* statistic was calculated for this range of *p*-values (z = 0.326 $p = 0.37$), indicating that they were not significantly different from *p*-values expected under the null hypothesis. In accordance with the pre-registration, this procedure was also re-run using multiply imputed values, also generating a non-significant result

(z = −0.115, p = 0.91). The same analysis completed using allocated intervention start dates did not result in materially different results (z = 0.314, p = 0.38).

Given the lack of overall treatment effect, further analysis of individual treatment response is unwarranted. In order to demonstrate the feasibility of such analysis we present the individual background characteristics of the ten 'high user' participants in **Table 4**.

Between Case Effect Size

The Between-Case Effect Size for the above data (n = 10), adjusted for small sample size, is 0.267 with a variance of 0.011 (see **Supplementary Appendix C** for sample code). This small effect size is consistent with the non-significant main finding. Studies have found that single case series often generate larger effects than those expected for group designs, and these effects vary widely depending on the technique used (Parker et al., 2005). In this context, the small effect size does not appear to be clinically meaningful.

DISCUSSION

The current study sought to describe and illustrate two powerful techniques for statistical analysis of Single Case Experimental Designs, which can be employed where the gold standard RCT may be difficult to implement. We used data from a brief intervention, which aimed to increase speech production skills in minimally verbal autistic children. The randomization test was used to compare the degree of improvement observed during the intervention period to the degree of change possible under the null hypothesis. This test indicated that results were consistent with the null hypothesis (no effect of intervention), with a corresponding small between-case effect size.

Although the intervention did not work as hoped, clearly the method has been useful and has provided insights into reasons why the intervention was not successful. An important factor that has become clear since the study was designed is the sheer volume of input and practice required to effect even a tiny change in expressive language in this population (e.g., Esch et al., 2009; Chenausky et al., 2016). The current study was limited by a 16-week timeframe that also included a baseline of a variable length, thus limiting the number of weeks of intervention. Future studies will require a longer time period to determine optimal treatment intensity and duration, and randomized case series with varying intervention periods are an ideal way to manipulate dosage and inform future larger scale trials.

A second key consideration for future replications is attrition. Our power analyses assumed a starting sample size (n = 18), however, only 10 children provided enough data for analysis, resulting in much lower power to detect statistically significant effects. Based on parent feedback, we expect that some attrition was related to frustration with technical difficulties. Due to the design of this study, those not engaging with the app could not be replaced. A major strength of this design is that it does not require baselines to be sequential; thus in future studies replacement could be used to manage attrition. Important considerations for future research also include specifying in pre-registration protocols how best to deal with missing data and adherence to intervention start date, in order to reduce bias in analysis.

The current study has laid useful groundwork for future replications in that we have demonstrated that an app can be used to elicit and record speech production attempts, and parents were able to accurately rate those attempts online following brief training. This means that one can have confidence in parent ratings, and they can be used to evaluate interventions, enhancing the scalability of this, and other apps. We also have an indication of how stable such attempts are in children who met criteria for minimal language, and what percentage of recruited families were able to meet the demands of the testing regime and comply with the intervention schedule. We have been able to illustrate individual differences in treatment response (**Figure 7**), and had we observed a meaningful treatment response we could have related this to individual child factors (**Table 4**). What we have demonstrated is that the chosen study design (multiple baseline with random assignment) and statistical approaches (Randomization Test and BCES) are feasible and straightforward to implement with real-world data, as generated by this sample of 10 participants. Based on our initial sample size and power calculations in **Supplementary Appendix B**, these methods are also more statistically robust than a comparable group study would be.

Randomized case series have a number of additional advantages. Firstly, they provide a much needed boost to power when compared with group designs, meaning that informative results can be obtained with fewer participants. This is critical for neurodevelopmental conditions that make obtaining a large and homogenous cohort challenging. Secondly, these designs are able to elucidate individual differences in treatment response, in a way that larger group studies cannot. Thirdly, case series are inherently a more feasible, low-cost, flexible endeavor, meaning they can be combined with clinical work and executed in a piecemeal fashion over a longer period. Finally, thanks to meta-analytic advances we can combine results from multiple case series in order to draw more robust conclusions about intervention efficacy.

CONCLUSION

The goal of this paper was to outline how to implement Single Case Experimental Design, by using random-assignment and the randomization test, as well as a between-case effect size to measure functional relationships between the introduction of an intervention and the outcome variable. The current study demonstrates that this is a robust method for rare, heterogeneous groups. While the BabbleBooster intervention did not lead to meaningful change in spoken language skills on this occasion, our goal is that this study will serve as a template for future studies that seek to answer a range of different therapeutic questions. Additionally, broader adoption of these methods will facilitate meta-analyses, allowing the

field to progress in understanding components of effective treatments for improving language in autism and other neurodevelopmental conditions.

The key take away points for any future students, researchers or clinicians seeking to adopt these methods are as follows: Firstly, plan for how many participants will be able to include, and how many times the dependent variable will be measured. These will likely be a function of funding or time constraints, and both have important implications for power. Within the overall study period, consider the minimum and maximum acceptable baseline periods. The maximum baseline will depend on participants' tolerance of repeated probes (boredom, irritability, practice effects) and the minimum intervention period is that which is expected to yield a meaningful intervention effect. A further planning issue is the number of probe items, how these are allocated and whether they include control items or randomization (see Howard, Best, and Nickels for further discussion of these issues). When it comes to selecting outcome measures, it is important to consider their reliability. In this study, we established parent/clinician reliability for coding speech attempts, which enhanced the scalability of the project by eliminating the need for the researcher to administer all test probes. Future studies will need to check the reliability of other combinations of delivery agents and language measures prior to data collection. Decide in advance how to handle missing data (how much missing data would exclude that participant's contribution?) or variations in adherence to intervention schedules. Finally, stability of the dependent variable is an important factor. If this is unknown and piloting is not feasible, power sensitivity analyses should take into account the impact of different correlations of the dependent variable at multiple testing points.

Ultimately, we would encourage clinicians and researchers to plan a study that is feasible for them, but to be realistic that they may not achieve adequate power in one "shot." However, if the studies are executed using the recommended techniques, alongside principles of reproducible open science, they are still valuable because they may be replicated at a later date by the same or different researchers. Lakens (2020) makes these points and adds that there is an ethical component to ensuring that the data we can feasibly collect is done in a way that leads to informative conclusions, either immediately or as part of subsequent meta-analysis. A huge challenge for the field is that RCTs are not always possible, yet single case studies alone are uninformative. However, by using the procedures outlined above we may be able to combine smaller studies through collaboration with other labs or clinics to yield informative conclusions, about intervention effectiveness and individual differences in treatment response.

DATA AVAILABILITY STATEMENT

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found below: https://osf.io/rzuwt/.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by UCL Research Ethics Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

JS had primary responsibility for study design, data collection, data analysis, and preparation of the manuscript. CN contributed to study design, oversaw data collection and data analysis, and provided detailed comments on drafts of the manuscript. Both authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

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Visual Sensory Experiences From the Viewpoint of Autistic Adults

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Although previous research has investigated altered sensory reactivity in autistic individuals, there has been no specific focus on visual sensory experiences, particularly in adults. Using qualitative methods, this study aimed to characterize autistic visual sensory symptoms, contextualize their impact and document any associated coping strategies. A total of 18 autistic adults took part in four focus groups which involved questions around visual experiences, the impact of these on daily life, and strategies for their reduction. Transcripts of each session were thematically analyzed allocating six key themes. Participants described a range of visual hypersensitivities, including to light, motion, patterns and particular colors, which contributed to distraction and were frequently part of a wider multisensory issue. Such experiences had significant negative impacts on personal wellbeing and daily life with participants describing fatigue, stress and hindrances on day-to-day activities (e.g., travel and social activities). However, the degree of understanding that participants had about their visual experiences influenced their emotional response, with greater understanding reducing concern. Participants employed a variety of coping strategies to overcome visual sensory experiences but with varied success. Discussions also highlighted that there may be a poor public understanding of sensory issues in autism affecting how well autistic individuals are able manage their sensory symptoms. In summary, autistic adults expressed significant concern about their visual experiences and there is a need to improve understanding of visual experiences on a personal and public level as well as for developing potential support.

Keywords: autism spectrum conditions, vision, visual sensory experiences, altered sensory reactivity, focus groups, qualitative methods, autistic adults, coping strategies

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INTRODUCTION

In addition to social interaction and communication difficulties, altered sensory reactivity, such as excessive (hyper-) or dampened (hypo-) sensitivity to stimuli, forms part of the autism diagnostic criteria (DSM-5: American Psychiatric Association, 2013; ICD-11: World Health Organization., 2019). Hypersensitivity describes an increased response such as extreme light or sound sensitivity

whereas hyposensitivity describes an obviously dampened response including apparent increased pain and temperature thresholds. In addition, individuals can also exhibit unusual interests in sensory aspects of the environment, such as excessive touching of object edges or fascination with reflections (Simmons et al., 2009; DSM-5: American Psychiatric Association, 2013).

Altered sensory reactivity is experienced by the majority of autistic people (Kientz and Dunn, 1997; Green et al., 2016). These experiences can be enjoyable or distressing (Smith and Sharp, 2013; Robertson and Simmons, 2015), and their magnitude has been found to be positively correlated with the number of autistic traits one may have (Robertson and Simmons, 2013). There is debate around whether altered sensory reactivity increases (Liss et al., 2006) or decreases (Kern et al., 2006) with age. Importantly, it remains throughout life (Crane et al., 2009) and affects each modality (Cléry et al., 2013; Baum et al., 2015) as well as multisensory processing (Marco et al., 2011; Beker et al., 2018).

The current study focused on visual sensory experiences of autistic adults. Informal discussions between autistic individuals and members of the research team, prior to this study, revealed the multidimensional difficulties that visual sensory experiences could cause for many autistic people, leading to them trying to manage these sensory issues themselves. This was exemplified by many autistic community members accessing unregulated "treatment" options such as tinted lenses which are claimed to be suitable management options[1] although there is no evidence base for this. Additionally, research has shown autistic individuals to present frequent ophthalmic conditions such as altered binocular vision, strabismus, refractive errors and compromised retinal structure (Little, 2018). It is possible that these conditions may be linked to autistic visual sensory experiences, but further research is needed. To be able to investigate this possible link, there is first a need to fully characterize visual sensory experiences together with their impacts on autistic people.

Bogdashina (2003) provided a list of visual hypersensitivity issues, such as focusing on fine detail and a dislike for extreme or flashing lights, and hyposensitivity issues, such as fascination of reflections or colorful objects and intensely focusing on objects or people. However, few studies have provided further characterization of visual sensory issues in autistic individuals, particularly in adults. Autistic visual hypersensitivities have been found to overlap with key characteristics of Meares-Irlen syndrome, also known as visual stress (Wilkins, 1995, 2003), defined as visual discomfort as a result of an increased sensitivity to repetitive patterns.

Subjective altered sensory reactivity in autism has been mostly explored using questionnaires. Findings from a recent meta-analysis of 55 questionnaire studies across children and adult populations (Ben-Sasson et al., 2019) supported the atypical nature of sensory symptoms in autistic individuals and highlighted the most consistent sensory experience was hypersensitivity. Whilst confirming heightened sensitivity across all sensory modalities (Tavassoli et al., 2014a,b), questionnaire studies have also made clear that the severity of sensory sensitivity varies between individuals (Ben-Sasson et al., 2008; Crane et al., 2009; Elwin et al., 2017). Of these studies, it is only Tavassoli et al. (2014a,b) who highlight the importance of investigating individual modalities so to not obscure intramodality differences. Specific to vision, they reported autistic adults to display heightened sensory sensitivity (Tavassoli et al., 2014a) and greater hypersensitivity (Tavassoli et al., 2014b) to visual stimuli relative to controls.

Quantitative research can be complemented and expanded upon by qualitative research. Although questionnaire methodology has provided extensive data about altered sensory reactivity in autistic individuals, it restricts the extent to which participants can express themselves and limits understanding of how experiences for each sensory modality may differ. Comparatively, qualitative techniques (e.g., focus groups or interviews) allow researchers to explore new ideas to greater depth and in different dimensions, such as attitudes, social interaction, thoughts and meaning (Malterud, 2001).

Qualitative studies have provided detailed evidence for general altered sensory reactivity in autistic individuals, although none have focused on visual sensory issues. Kirby et al. (2015) used semi-structured interviews to investigate sensory experiences in autistic children. Experiences were generally described as "likes or dislikes" with interviewers unable to determine sensory issues within individual modalities. They concluded this to indicate that autistic children view their experiences as multisensory. Altered sensory sensitivity amongst autistic adults has been documented using semi-structured interviews (Smith and Sharp, 2013), focus groups (Robertson and Simmons, 2015), and analysis of personal accounts (Jones et al., 2003), but this was not explored for individual modalities. However, some visual experiences were superficially reported including difficulties tolerating a range of stimuli such as bright environments, artificial lighting, patterns, unpredictable movements, visual distractions, fine detail, and particular colors (Jones et al., 2003; Smith and Sharp, 2013; Robertson and Simmons, 2015). Child group interviews by Robertson (2012) revealed similar visual difficulties; some colors, bright lights and screens, and additionally certain shapes can cause painful sensations.

The impact of general altered sensory reactivity on autistic individuals has previously been investigated revealing negative and pleasurable emotions, negative physical symptoms, effects on attention, and both positive and negative impacts on daily living (Jones et al., 2003; Smith and Sharp, 2013; Robertson and Simmons, 2015). Strategies to cope with these include purposeful exposure to positive stimuli or avoiding, accommodating, distracting away from, and seeking the positive aspects in negative stimuli (Jones et al., 2003; Smith and Sharp, 2013; Robertson and Simmons, 2015). In an interview study about autistic adults' daily lives, Robledo et al. (2012) found participants to mention that although visual stimuli could cause negative emotions and physiological responses, certain lighting or color combinations could be enjoyable.

While these qualitative studies have provided a superficial description of visual experiences, there are no studies that have specifically examined autistic visual sensory experiences in depth. General findings regarding impacts of sensory experiences on

[1] https://www.read123.co.uk/en/the-use-of-color-therapy-and-colored-lenses-in-autism/

quality of life and coping strategies cannot be assumed to apply across every sense. Studies which have documented subjective visual experiences (Jones et al., 2003; Robertson, 2012; Robledo et al., 2012; Smith and Sharp, 2013; Robertson and Simmons, 2015) have not attempted to explore these further or characterize them in-depth but instead summarize them broadly alongside other modalities.

On the other hand, a large body of work has examined vision using cognitive and psychophysical tasks in autistic people (Simmons et al., 2009; Schauder and Bennetto, 2016; Apicella et al., 2020; Federici et al., 2020). Various studies have investigated performance of autistic and non-autistic individuals in tasks linked to early visual processing, such as visual acuity (Tavassoli et al., 2011; Albrecht et al., 2014; Tebartz van Elst et al., 2015) and contrast sensitivity (Koh et al., 2010). Higher level visual processing has also been explored, for example, face recognition (Tang et al., 2015) and global (Van der Hallen et al., 2015) and biological motion perception (Todorova et al., 2019). While findings are mixed, these studies suggest fewer group differences for lower than higher level visual processing. Autistic people have also been found to exhibit perceptual differences, with a superiority or preference in processing local compared to global information (Plaisted et al., 1999; Rinehart et al., 2000; Happé and Frith, 2006; Mottron et al., 2006; Simmons et al., 2009; Muth et al., 2014; Kabatas et al., 2015).

It is evident that whilst qualitative studies have provided detailed evidence for general altered sensory sensitivity in autistic individuals, the visual sense has not been explored to the same extent with no previous studies focusing specifically on subjective visual sensory experiences. Moreover, these studies have been conducted mainly from a psychology or psychophysics point of view; how would a vision and ocular health expert interpret these findings? This is an important gap to fill as improved descriptions of visual sensory issues, as well as from a different professional perspective, can suggest directions for future quantitative studies. For example, if descriptions of autistic visual sensory issues overlap with characteristics of visual stress, binocular vision symptoms, poorly corrected refractive error or are more suggestive of cognitive mechanisms (Happé and Frith, 2006; Mottron et al., 2006; Van de Cruys et al., 2014), future work can be targeted to test these links. Additionally, it is clear from previous reviews (Schauder and Bennetto, 2016; Ben-Sasson et al., 2019) that there has been a greater focus on investigating sensory difficulties in autistic children than adults. In Ben-Sasson et al.'s (2019) meta-analysis, only 7% of questionnaire studies that examined sensory symptoms involved adults. Overall, a detailed characterization of the multi-faceted visual sensory experiences in autistic adults, the specific impacts of these on daily life and the strategies employed to cope with these does not exist. The aim of the current study was to gain a detailed insight into the everyday visual experiences of autistic adults along with their impact and coping strategies employed, from the point of view of an optometrist.

A qualitative approach was taken in order to explore the full range of visual experiences that autistic people report and to what extent these impact their daily lives. Focus groups were employed as they allow opinions to be collated from a relatively larger sample, compared to one-to-one interviews, and have been successfully conducted with autistic adults in previous research (Robertson and Simmons, 2015; John et al., 2017; Koffer Miller et al., 2017; Gowen et al., 2019). Furthermore, interactions between members in a focus group allow researchers to understand the range of opinions as well as the level of agreement about topics (Barbour, 2008), particularly suitable for the current study's aims.

MATERIALS AND METHODS

Recruitment and Participants

An advert was publicized by email and social media using the Autism@Manchester network, local autism groups and the university platforms. Flyers were also displayed around the university campus and handed out at autism events. Inclusion criteria were (i) being formally diagnosed as autistic; (ii) absence of a learning disability; (iii) aged 18 years or above; (iv) being able to travel to the university, and; (v) availability to attend one of the specified focus group sessions.

An opportunity sample was recruited for this study. Although 27 participants signed up to a focus group session, nine did not attend. A total of 18 autistic adults took part, aged 25 to 67 years (mean age 47.1 years), of which six were female. All had a formal diagnosis of an autism spectrum condition (autism/Asperger's syndrome/ASC) visually confirmed by a diagnosis letter, and were from the northern regions of England. In terms of ocular history:

- 17 participants presented with at least one ophthalmological condition;
- 16 participants wore a refractive correction;
- 8 participants had an additional eye condition including amblyopia, visual stress, keratoconus, light sensitivity, Graves Ophthalmopathy and history of an eye trauma;
- 2 participants had undergone eye surgery such as cataract extraction, removal of a corneal ulcer or laser vision correction;
- 4 participants had received eye treatment such as use of eye drops or eye patching in childhood.

This study received ethical approval from The University of Manchester's Research Ethics Committee (2019-6025-9932) and participants provided informed consent.

Study Development and Procedure

The research team comprised KP, a Ph.D. student with training in qualitative methods and practicing optometrist by profession; EG, a researcher in the field of sensory perception and motor control in autism; CD, a professor of clinical optometry with a specialist interest in helping those with uncorrectable visual impairment; and CP, a senior lecturer in optometry as well as practicing optometrist with a specialist interest in binocular vision. Across the team existed a wealth of knowledge about the visual system, refraction and ocular health which allowed the research to take a unique approach, as opposed to previous research which has taken a more psychological stance.

The design and procedure of this study were developed in collaboration with the Autism@Manchester Expert by Experience Advisory Group[2]. Thereafter, the research team worked closely with two adult autistic advisors (JP and PB) who ensured an appropriate protocol for the focus groups which would be autism-friendly.

A total of four focus groups were held as this number can reveal up to 90% of all themes (Guest et al., 2017). Each contained four to six participants. Participants were randomly allocated to a focus group depending on their availability to attend. Prior to attendance, participants were sent a "what to expect during the study" document (SM 1) to prepare them for their visit. Upon arrival, they were taken to the focus group room and offered refreshments whilst written consent was taken. Thereafter, they completed a questionnaire which collected basic demographic and diagnosis information as reported above. The focus groups were facilitated by one member of the research team (KP) who followed a predetermined schedule (SM 2). Participants were fully aware that they had access to a quiet room and were able to leave the discussion at any time without having to give a reason. They were also reassured that the data collected during the focus group would be pseudonymized. Another member of the research team (EG or CD) was present to assist with running the sessions which ran for 1-2 h, excluding a short break midway.

In line with recommendations from Durand and Chantler (2014), four key questions were presented to the groups of which three are explored in this paper:

> Q1. Does anybody feel they experience any visual problems or unusual visual symptoms?
> Q2. Do you feel you can do anything to improve these symptoms?
> Q3. How do your visual issues impact your daily routine?

The remaining question (Q4) "what are your experiences of an eye examination?" was unrelated to the topics explored in this paper and will be discussed in a future article. Q1 allowed the researchers to explore the range and magnitude of autistic adults' visual experiences. A key aim of this study was to characterize these experiences in detail by understanding what steps autistic adults take to tackle these (Q2) and what affect they have on an autistic adults' life (Q3).

Data Analysis

The focus groups were audio recorded and then transcribed, with participants pseudonymized, by an external university approved service for intelligent verbatim transcription. Transcripts were thematically analyzed to allow the broad range of data to be brought into meaningful themes. Compared to other qualitative analysis methods, thematic analysis allows data sets to be richly described as a whole and goes further than just summarizing data (Braun and Clarke, 2006; Maguire and Delahunt, 2017). The analysis aimed to be exploratory and the research student (KP) took an inductive, semantic and realist approach from the point of a non-autistic optometrist.

[2]www.autism.manchester.ac.uk/connect/expert-by-experience

The Braun and Clarke six-step technique (Braun and Clarke, 2006) was followed as this framework is flexible and can be easily applied to a variety of research questions. Firstly, the accuracy of each transcript was checked against the original recordings. The research student then familiarized himself with the data by re-reading through the transcripts whilst making any initial notes of key ideas. The second phase involved re-reading and line-by-line coding of the transcripts to identify features (words, sentences or paragraphs) of the data related to the scope of the study. This was done by hand and codes were written on sticky notes.

In the third phase, codes were grouped to form initial themes. For this, as per the recommendations of Braun and Clarke (2006), a physical thematic map was created by arranging the sticky notes according to similarity in content or ideas. This allowed the research student to visualize the formation of higher-level themes. These three stages were followed for each transcript and moderate alterations were made to the thematic map as more transcripts were analyzed.

Data saturation was reached with no new themes developing from the fourth focus group. The fourth phase reviewed the allocated themes against the dataset as a whole. It was important that the themes captured all relevant aspects of the data. The themes and codes were discussed amongst the research team (KP, CD, CP, and EG) to improve the rigor of the analysis and ensure a valid interpretation of the data. The team agreed that the codes summarized the relevant aspects of the data well, however, some themes could be grouped together as they were (a) very small and (b) closely related. The thematic map was reorganized as per these modifications (see **Table 1**).

Themes were appropriately named and given a short definition in the fifth phase. The research team had to ensure that a theme's name gave an immediate reflection of what was covered therein and highlighted its relevance to the scope of the study. Additionally, a detailed analysis of each theme, most either complex or large, lead to the allocation of multiple subthemes. The final phase involved bringing together the themes and supporting data in a report. For this, appropriate quotes were chosen from the data set which justified the research findings (see section "Results"), and the overall outcomes needed to be discussed in the context of the study aims and existing literature (see section "Discussion").

RESULTS

A final six themes were allocated to the data and are listed in **Table 1** under the question from which they arose; themes, theme definitions and corresponding subthemes are presented.

The remainder of this section describes these themes in further detail. The participants are referred to by a number (P1-18).

Theme 1: Altered Visual Experiences

Visual Hypersensitivities

Participants described a variety of issues, relating to visual hypersensitivity, which refers to an increased sensory sensitivity rather than threshold detection sensitivity. Hypersensitivity to lighting had multiple aspects; bright, flickering, fluorescent,

TABLE 1 | The six allocated themes and their definitions as well as respective subthemes, grouped according to the question from which they arose.

Themes	***Theme definition***	***Sub-themes***
Question 1: Does anybody feel they experience any visual problems or unusual visual symptoms?		
Altered visual experiences	Visual symptoms or unusual occurrences experienced by participants	Visual hypersensitivities Eye movements Visual experiences vary from person to person
Autistic individuals' vision-related knowledge	The level of understanding participants had surrounding their vision and ocular health, and the impact of this	Degree of awareness Impact of awareness
Question 2: Do you feel you can do anything to improve these symptoms?		
Coping strategies	Methods adopted by participants to tackle their visual experiences	Avoiding visual clutter Optical correction choices Colored overlays/lenses Lighting alterations Just cope with it A multisensory experience
Question 3: How do your visual issues impact your daily routine?		
Impact on personal wellbeing	The multi-dimensional impact of visual experiences on participant wellbeing	Physical wellbeing Mental wellbeing Emotional wellbeing
Impact on daily life	The impact of visual experiences on participants' daily lives	Home life Work life Public places Travel Social life
A poor public understanding of sensory issues in autism	The perception of a poor awareness in the general population surrounding autism and the participants' reaction to this	

"strip," and "spot" lighting caused discomfort. Participants, with and without a diagnosis of visual stress, described difficulties reading and viewing certain patterns. P4, who suffered with visual stress, said, *"...it's like, the letters... flicker around the edges sometimes. As if the letters are bleaching into the gray bits..."* which describes typical visual stress characteristics. Our participants also portrayed visual stress to be caused by day-to-day striped visual images such as "grills on buildings" or "radiators." P9 explained an adverse response to patterns, again characteristic of visual stress, although they did not have a formal diagnosis:

> *"...I definitely sometimes have what feels like a physiological response to the patterns...It's like you've been punched in the stomach. It's a real strong emotion that comes over you like when there's loud noise."*

Hypersensitivity to particular colors could cause an adverse response, *"...I get like a physical reaction to them as though they've hit you"* (P9), and was suggested to largely influence our participants' likes and dislikes, where they visited and what they selected. *"I mean I really don't like the color yellow. I mean the railing down there is toddler screaming levels of irritation. And I'm not fond of bright reds"* (P8); *"...all of my upholstery I've chosen, it's beige... But for me, I mean it's not that nice to look at... But if I had any bright colors then I would just avoid that room"* (P11). Participants indicated that the impact of color cannot be predicted because the contrast with the surroundings, the pattern formed with other colors and the combination of colors are all influential factors.

Hypersensitivity to visual motion occurred mostly in crowded, busy environments and was implied to be due to a combination of visual clutter and movement. P14 explained:

> *"It's the movement of other people, because to me it's...if I'm in my living room, my children are running back and forth across, I find that very stressful. They need to be on one side, not going across my field of view all the time. I get stressed and angry."*

The main impact of visual hypersensitivity was distraction. P1 said:

> *"...the general theme of being distracted by visual things is an issue for me. Especially because I wear glasses and they always get dirt on; I notice the dirt a lot more than other people."*

Participants suggested they were more aware of their full field of view and had difficulty paying attention to a particular part of it. A conscious effort has to be made to overcome distraction, *"I'm not at the moment looking the other way, but if I was tired, I'd be much more distracted. So, it's almost like playing a filter to try and filter it all consciously"* (P5).

The inability to overcome distraction caused negative emotional responses such as anger and frustration, *"...it drives me mad..."* (P5). However, not all distractions had a negative impact. Some participants said they occasionally dedicated their attention to one visual stimulus which fully occupied their sensory system to prevent distraction from other stimuli, *"...so I can ignore the sounds and the visual stimulus of people talking, I focus as firmly as I can on what I'm reading"* (P8).

Eye Movements

Four participants expressed difficulties with controlling eye movements. Others had been made aware of problems with eye tracking through research studies requiring eye calibration which suggests that these issues are not always apparent to autistic individuals. P1 reported, *"My eye tracking is a bit weird. I think I don't look at...the thing I'm intending to look at sometimes or following it correctly"* and confirmed this to be the case *"...all the time to some extent."* P13, who did not declare any binocular vision problems, stated:

> *"...I hadn't realized how much I'd struggled [with reading] for years, because it's sort of double, but it's very subtly double. And sometimes it just goes like that [hand gesture indicating diplopia]."*

Visual Experiences Vary From Person to Person

Participants' visual experiences varied from person to person depending on the severity of their altered visual sensory sensitivity. For example, P7 said:

"...I have more issues with sound. Not excessively, as sometimes you hear about, but certainly more so with sound than with vision. Which is why, like I say, I couldn't really relate to what was being discussed..."

Many participants felt they had good vision and could *"see clearly"* (P14), however, some felt their eyesight was genuinely poor. P15 said, *"...I never feel I can see well enough, ever."* Participants were quick to suggest that they could differentiate perceptual symptoms from sight issues, *"I don't think my eyes see differently from other people, I think I process it differently"* (P1).

Theme 2: Autistic Individuals' Vision-Related Knowledge

Degree of Awareness

The degree of awareness about vision and eye health varied across the participants. They expressed little flexibility with their definition of good vision, thinking 20/20 acuity and a low spectacle prescription defined this. The presence of non-pathological floaters, the time taken for light adaptation, and foveal bleaching as a result of viewing a bright object all seemed to be perceived as poor eye performance by participants although these are normal physiological phenomena, *"So the floaters are the big thing... At work, the dark floaters at work, that's not good vision"* (P6).

Impact of Awareness

The degree of understanding that participants had about these aspects in turn impacted their emotional response to them. In cases where there seemed to be a lack of understanding, participants expressed negative emotions such as fear, anxiety and feeling abnormal, "*I just feel like I'm made up of bad code*" (P6). Well informed participants appeared to be less concerned about the same experiences.

Theme 3: Coping Strategies

Avoiding Visual Clutter

Participants indicated that visually busy environments overwhelmed their sensory system, so tended to avoid them. P3 said, *"...the city center, being surrounded by buildings and people like it's all too much visual information."* Where avoidance was not possible participants minimized the time they had to experience visual clutter, *"I tend to use the same shops. So I know exactly where things were last time, barring the usual rearrangements. So I try and get it done quickly. So I can get out of there*" (P8).

Optical Correction Choices

Interestingly, participants discussed their choice of spectacles. P1 said, *"different glasses would help me, like if they were rimless or I wasn't distracted by a frame..."* Although different participants interpreted the effect of the frame size in different ways, the general conclusion was to avoid seeing the rim, *"you're physically aware of them, this is slightly annoying"* (P7). Participants requested more reassurance when being dispensed new spectacles and identified the need for a relatively longer adaptation period to these.

Although sunglasses and photochromic lenses are helpful as they reduce light levels, they were suggested to be only a partial solution. P8 stated, *"...it's a little disconcerting having that artificial darkening when I'm used to things seeming bright,"* which leads to the issue of feeling *"detached"* (P10). It appears that this population may struggle with optimal light levels without visual sensory issues being compromising.

Colored Overlays/Lenses

The use of colored overlays and lenses was beneficial for some participants who experience visual stress symptoms. P13 stated, *"...my reading speed doubled...I could actually see properly."* P2 said, *"...I never saw the social world, I never saw people, never saw expression..."* prior to using colored lenses. However, they were not equally beneficial for all. P14 said her tinted lenses were *"...not all that helpful with the visual stress. Just good with headlights and blueness."*

Lighting Alterations

Light alterations increased participants' ability to cope in artificially lit environments. Reducing light levels can improve visual ability, *"...I can see very well in low light"* (P8). Whereas natural color temperatures, such as 'daylight' ease visual symptoms and are *"preferred"* (P1), warmer color temperatures are *"very warm, very comforting"* (P4). Some participants also suggested that blue blocking lenses *"relax"* (P10) them and make spectacles feel better.

Just Cope With It

The final approach was to *"cope, cope as best as you can"* (P9). Participants described that as a hypersensitivity is increasingly provoked it induces a greater negative emotional response, leading to growing distress. Apart from being stressed and anxious, they are likely to inconvenience themselves by trying to prepare for every situation, *"I just carry loads of different types of glasses with me... So it just means I'm covered for all eventualities..."* (P13).

A Multisensory Experience

It was challenging for some participants to think about vision-related coping strategies because visual sensory symptoms usually occurred as part of a multisensory experience for them:

"...it's difficult to pull out that that is due to any particular reason really. I mean the talking to people I struggle with if there's the stripy shirt distraction issue, because it's something that's grabbing your attention away from what they're saying. At the same point as there's five different people behind them whose conversations you're listening to at the same time because you can't screen them out. So it's difficult, isn't it, to say what is due to which issue." (P9).

Theme 4: Impact on Personal Wellbeing

Altered visual sensory reactivity had multiple impacts on our participants' wellbeing.

Physical Wellbeing

Physically, visual experiences are *"a gradually fatiguing thing"* (P10), which impacted the participants' functionality. P14 said, *"flickering lights, like the sun behind trees, makes me sleepy."*

Additionally, many participants expressed sleeping difficulties, especially during summer months due to longer daylight hours.

Mental Wellbeing

Participants lacked a feeling of self-worth due to low self-confidence as expressed by P6, *"I feel like my genetics are just really bad codes, just full of defects and errors. And that's [referring to their vision] just another error to catalog."* P4 suggested that this lack of confidence could be a result of not knowing *"what other people see"* which constantly makes them doubt if they are seeing the world in the same way as non-autistic individuals.

Emotional Wellbeing

Emotional wellbeing varied amongst our participants. P1 said, *"it [visual hypersensitivity] just makes me generally stressed all the time and less able to deal with other things."* P6 was angry about his vision, *"I'm not happy with my eyesight at all. No, I'm not,"* whereas P12 found his vision simply *"overwhelming."* Conversely, participants who saw the advantages of their visual experiences portrayed themselves as relatively more positive in the way they spoke, their responses and their body language, as explored in the next theme.

Theme 5: Impact on Daily Life

Home Life

Visual experiences were a hindrance in home life for some of our participants, especially for tasks requiring concentration such as *"cooking and sewing"* (P1); these can be difficult to complete with ease and enjoyment. Some participants, however, saw their visual hypersensitivities as an advantage especially for hobbies. P4 stated:

> *"...seeing details is a double-edged sword. On the one hand you could get overwhelmed with all the detail. But at the same time, it also means... When I'm painting, I can see far more detail than other people can see. I can spot things that other people miss."*

Work Life

Regarding employment, participants expressed positive views. In particular, sensitivity to fine detail was an advantage at work:

> *"...I just use my ability to pay attention to visual detail more, more in my professional work which involves a lot of image analysis and data processing to produce those images..."* (P2).

Public Places

Having to avoid certain environments due to visual hypersensitivities meant participants were more likely to stay at home. They expressed issues with cinemas, large shops, hospitals and lecture theaters specifically. However, issues with public places cannot solely be blamed on visual experiences. Participants recognized that these difficulties are more due to multisensory problems and anxiety which collectively overwhelms them:

> *"it's a multi-level thing. I mean hard, easy to clean floors, which means every person stepping around is bang, bang, bang. You've got people moving around randomly. You've got the bright lights. and you've got all the people talking simultaneously."* (P8).

Travel

Participants said they can be distracted and overwhelmed by visual clutter, headlights and objects that catch their attention when driving. Some had given up driving due to their visual experiences, whereas others had not pursued driving due to a fear of these:

> *"I mean headlights are a problem when driving in the dark. It's like the headlights seem to bleed and wash out some of the rest of the visual experience, and you need that data when you're driving in the dark."* (P10).

Participants found it difficult to use public transport due to the artificial lighting at night or sunshine on bright days. The majority of participants in our study suggested that the issues with public transport were multidimensional and again could not be accounted for just with vision:

> *"...dealing with the driver, walking past all the people to find a seat and finding a seat with the person next to me... And then just the overall noise and the rattling of the engine and the windows"* (P10).

Social Life

Many participants felt that sensory experiences contributed to difficulties in their social life. When asked about inclination to attend social events, P4 said, *"I think less inclined purely because I don't want the overload of all the sensory input."* As described by P1, the stress induced by visual experiences reduces their ability to deal with other situations such as *"interacting with people."* Due to these experiences, P3 felt limited in her social life:

> *"I drive because I can't do public transport. And so, if we think about the impact socially and stuff then a lot. So, I can't go out drinking because I have to drive home and stuff like that as well."*

However, our participants' difficulties and limitations in social situations could be misinterpreted as being *"antisocial"* (P4).

Theme 6: A Poor Public Understanding of Sensory Issues in Autism

Participants described a lack of awareness in the general population regarding the sensory difficulties autistic adults face. P5 felt the ignorance of some non-autistic individuals toward autistic people is *"absolutely disgusting."*

Educating the general public about the sensory issues in autism is important to heighten understanding, *"... now my wife understands why I have to leave things early. Before she just thought I was being antisocial. Now she knows she's more understanding about it"* (P4).

There was also a fear amongst our participants that their difficulties may not be understood by employers or public services. For example, P10 said:

> *"...I'm really dreading I think to have some kind of sensory conversation with an employer. Because there's some environments I go into now, like the hospital, and I don't think I could physically tolerate that. So yeah, I think this has big implications..."*

DISCUSSION

The current study is the first to provide an in-depth qualitative investigation of autistic visual experiences, together with their impacts on daily life and coping strategies. A total of 18 autistic adults, without learning disabilities, attended a focus group meeting at The University of Manchester. The opinions of these participants were elicited to gain a holistic understanding of the visual experiences of the autistic adult population. It builds on previous work which has briefly documented visual issues in the context of a broader study on altered sensory reactivity (Jones et al., 2003; Robertson, 2012; Robledo et al., 2012; Smith and Sharp, 2013; Robertson and Simmons, 2015), and highlights significant concerns amongst autistic adults regarding their vision, visual sensory experiences and the impacts these have.

Characteristics of Visual Issues

As noted by previous literature, our participants highlighted increased sensitivity to different aspects of lighting (Bogdashina, 2003; Leekam et al., 2007; Robledo et al., 2012) and fine detail (Simmons et al., 2009; Kabatas et al., 2015). Participants discussed strong likes and dislikes for particular colors, agreeing with the findings of a case report by Ludlow and Wilkins (2009). This also appears to be analogous to the outcomes of Grandgeorge and Masataka (2016) who investigated color preference in autistic boys aged between 4 and 17 years, finding they were significantly less likely to prefer yellow, compared to age-matched controls, but more likely to prefer green and browns. Such color preferences were suggested to be a result of autistic visual hypersensitivities, also indicated by our participants.

Additionally, participants reported key symptoms of visual stress when viewing repetitive patterns, including flickering, fading and a strong discomfort (Evans and Stevenson, 2008). Although a few participants described the impact of this phenomena particularly with reading, it was largely discussed as a global experience impacting several aspects of daily life, and dependent on the combination of colors which produced an uncomfortable contrast. Robertson and Simmons (2015) also found their focus group members to describe visual stress symptoms in the context of more global aspects such as the pattern formed by the layout of products on shelves in a shop. In view of this and other studies reporting first-hand accounts of pattern sensitivity in autistic adults, future work should investigate whether the visual experiences of autistic people are at all related to Meares-Irlen syndrome.

Our participants suggested issues with eye tracking, visual location and control of binocular vision (reporting diplopia), but it is unclear how these reports would relate to formal laboratory measurements reported in the literature. A review and meta-analysis by Johnson et al. (2016) reported that autistic individuals can have altered eye movements, specifically poor eye tracking, impaired saccade inhibition and saccade dysmetria, but do not have difficulty initiating saccades or engaging/disengaging from targets. Additionally, studies have found autistic people to make faster eye movements during predictive saccade tasks (D'Cruz et al., 2009; Kovarski et al., 2019).

Autistic individuals are more likely to develop ophthalmological conditions (Little, 2018). These include refractive error, binocular vision and ocular muscle balance anomalies, and altered retinal structure. It is not known whether any of these deficits may contribute to autistic sensory symptoms although this would be a valuable relationship to investigate.

It is important to note that visual sensory experiences varied amongst our participants, depending on how sensitive they were to their vision. For example, those more sensitive to sound did not necessarily fully relate to the accounts of participants reporting severe visual symptoms. This variability is evident in existing research on autistic altered sensory reactivity and highlights the importance of not merely generalizing findings across the autistic adult population.

Visual Sensory Experiences and Attention

Autistic individuals have displayed and described impairments with attention (Liss et al., 2006); Patten and Watson (2011) discussed alterations in three broad features of attention in autistic children: orientating, sustaining and shifting. Attention is closely linked with distraction and previous studies have demonstrated that autistic individuals have difficulty ignoring irrelevant distracting sensory information (Christ et al., 2011; Adams and Jarrold, 2012; Smith and Sharp, 2013). Reasons for this could be greater perceptual capacity (Remington et al., 2009; Bayliss and Kritikos, 2011; Tillmann and Swettenham, 2017) or enhanced sensory sensitivity (Liss et al., 2006), both of which have been found to be positively correlated to each other in a recent study by Brinkert and Remington (2020). Our participants indicated that they had to make a conscious effort to attend to their central field of view and ignore their peripheral vision, in agreement with Mottron et al. (2007) who investigated lateral glances in autism.

Difficulties with distraction led to issues with driving for our participants. The visual sensory experiences encountered during driving can be overwhelming and autistic adults can struggle to pay attention where it is required. The literature suggests this population display relatively more problematic driving behaviors (Daly et al., 2014), and are less likely to attend to all relevant parts of their visual field during driving. However, visual issues can be one of many aspects which impact driving ability (Reimer et al., 2013). Autistic individuals are more prone to becoming anxious (Reimer et al., 2013), and have shown difficulties with motor coordination, staying in lane, control of speed, and adapting to unexpected situations during driving (Classen et al., 2013).

Multiple Impacts of Visual Sensory Experiences

Our study suggests that visual experiences can contribute to difficulties maintaining emotional, mental and also physical wellbeing. As well as causing pain and negative physiological responses, fatigue caused by altered visual sensory reactivity appeared to directly impact on the functioning of our participants. Emotionally and mentally, our participants largely expressed low mood and negative feelings, such as fear and

stress, due to their visual experiences. They saw themselves as excluded because of the sensory problems they faced, which included vision.

Visual experiences could contribute to poorer daily living skills that are present in autistic individuals (Smith et al., 2012; Bal et al., 2015). Chores in the household such as cooking, and visiting public places including shops and hospitals, were all made more difficult as a result of visual experiences for our participants. They suggested being put off tasks which demand a lot of visual attention; visual experiences limit them to a few tasks which they can complete and enjoy.

However, sensory experiences can have positive aspects too. They can be enjoyable specifically when they or the associated anxieties are under control (Jones et al., 2003). Our study found that hypersensitivity to fine detail can prove an advantage to autistic people as they can detect details which non-autistic individuals may overlook. Although this was the case, participants did not mention this to be related to any positive effect on their mental or emotional state as also noted by Robertson and Simmons (2015). Robledo et al. (2012) also observed that some participants enjoyed visual stimulations such as bright lights and particular color combinations. Seeking the positives in sensory experiences was identified as a coping strategy by Jones et al. (2003).

Coping With Visual Sensory Experiences

Little research has been carried out to date to investigate autistic individuals' coping strategies for their sensory issues (Jones et al., 2003; Smith and Sharp, 2013; Robertson and Simmons, 2015). Our results agree with Smith and Sharp (2013) who found that moderating factors, such as reduced sensory inputs, reduced sensory intensity, predictable environments and the autistic person being calm, can lower the impact of otherwise overwhelming sensory experiences. Our participants suggested that autistic adults can feel overwhelmed by a large variety of visual information. They attempted to prevent sensory overload by means such as avoiding visually cluttered public places at peak times and shopping at the same stores as they would know where items are kept.

The effort made by participants to prevent sensory overload by avoiding social interaction could be misunderstood as awkwardness or being uncooperative. Participants were disappointed and anxious about the poor public understanding of autism and associated sensory issues. They agreed that this has to be improved, which agrees with recent recommendations in The Autism Dividend report (Lemmi et al., 2017). Our study has attempted to describe the visual experiences of autistic adults without learning disabilities so that professionals, service providers and members of the public can develop an understanding of this and be more accommodating.

Altering lighting, in terms of brightness and color temperature, was also beneficial for our participants and felt to improve visual performance. This could be related to visual stress with these light adaptations having a similar effect to the use of colored overlays or lenses. Participants also commented on the relaxing nature of blue blocking lenses, agreeing with a randomized trial in non-autistic individuals (Kimberly and James, 2009) which concluded that these can significantly improve mood.

The benefits of colored overlays or lenses in autism have been speculated upon. Some studies have shown improved reading speed (Ludlow et al., 2008; Ludlow and Wilkins, 2009), better control of behavior, coordination and personal space (Ludlow and Wilkins, 2009), and improved ability to characterize the intensity of facial expressions (Whitaker et al., 2016). Some of these social aspects were confirmed by the personal accounts of our participants. Though colored lenses reduced visual stress for some of our participants, for others they did not, and no rigorous controlled trials have yet been conducted in this area. It is therefore crucial for optometrists and autistic individuals to know that while there may be possible benefits of prescribing colored lenses, they may not work as expected in all instances and further research is needed.

The final approach to "just cope" resulted in participants experiencing a variety of negative emotions. As per Carver et al. (1989), there are two distinct forms of coping: problem focused and emotion focused coping. In terms of our findings, although participants' coping strategies could be grouped as one or the other, we do not know how they reached these stages.

Some of these coping strategies could be underpinned by "compensatory mechanisms," which involves alternative cognition to bypass cognitive difficulties. As a result, autistic people display fewer behavioral symptoms despite continued underlying cognitive and neural deficits. These mechanisms can be applied to compensate for particular cognitive atypicalities, as opposed to "camouflaging" which aims to mask all autistic traits (Livingston and Happé, 2017; Livingston et al., 2019). It is therefore not surprising that Robertson and Simmons (2015) suggest specific coping strategies developed by autistic adults could help explain some of the unusual behaviors adopted by this population. The overall message from our results is that visual experiences result in a variety of issues for autistic adults which result in strong positive and negative emotional impacts. A range of coping strategies are employed to deal with these.

Participant Interpretations of Visual Sensory Experiences

Our participants thought that many of their visual experiences were a result of higher-level processing issues and not necessarily due to uncorrected refractive error or poor binocular vision. However, the degree of vision-related knowledge varied amongst our participants and appeared to influence anxiety about their visual issues. For example, some were worried, feeling normal ocular phenomena were a sign of poor eye performance while the opposite was true for those who had a good understanding of phenomena such as light adaptation and floaters. It is important to note that health-related anxiety is likely to vary similarly in the general population too so we cannot conclude that this is an issue confined to autistic adults. Nevertheless, to reduce this anxiety in autistic individuals, there is a need to increase their understanding around vision and eye-health.

Moreover, participants indicated that their visual experiences usually occur as part of a larger multisensory experience which may be a reason as to why they generally found it difficult to specify the contribution of vision to their sensory experiences. Issues with public transport are a good example of this; hypersensitivity to light is one aspect, but this is part of a multisensory issue alongside anxiety. Indeed, processing multisensory stimuli is altered in autism (Robertson and Baron-Cohen, 2017). It may be the case that altered sensory processing in one modality has an impact on other modalities. This could amplify or dampen the sensory symptoms.

Limitations and Considerations

To our knowledge, this is the first in-depth qualitative study which set out to explore the subjective visual issues experienced by autistic adults, the impacts these have on their daily lives and what they do to minimize these. However, our results can only be considered for autistic adults without learning disabilities. Those with coexisting learning disabilities or other neurodevelopmental disorders may also experience visual sensory symptoms: an observational study for individuals who cannot express their symptoms verbally may identify corresponding behavioral signs. Additionally, participants were limited to those who could communicate in a focus group setting. A further study offering interviews or online focus groups for individuals who cannot take part in a physical verbal group discussion may have yielded further insights. Individuals may have been more likely to participate in this study if they were aware of having visual problems which could have resulted in reports of more negative or extreme experiences. However, as our aim was to describe visual experiences rather than quantify them this has less of an impact on our results.

Sample size determination is difficult in qualitative research and there are alternative approaches suggested for this. A recent article by Braun and Clarke (2019) discusses data saturation in the context of thematic analysis and suggests that it is difficult to justify sample size with data saturation for studies which aim to be exploratory, inductive and that do not ask exactly the same questions during every focus group. In our study, recommendations regarding number of focus groups by Guest et al. (2017) and data saturation, during the planning and data analysis phases respectively, were used to confirm a suitable sample size. However, in line with suggestions by Braun and Clarke (2019), our focus groups were on a very select topic and all of our participants were autistic and had experience of an eye examination, therefore each was likely to have more "information power" (Malterud et al., 2016), meaning our modest sample size was acceptable.

Many of our participants also had coexisting conditions such as dyspraxia, ADHD and anxiety disorder which may have influenced our results. There is evidence that individuals with dyspraxia have defective global spatial processing (O'Brien et al., 2002). Mogg et al. (2000) concluded that individuals with generalized anxiety disorder display altered eye movements to threatening facial expressions. In a national United States investigation, there was a greater prevalence of ADHD among children with visual problems which could not be corrected with spectacles or contact lenses (DeCarlo et al., 2016). Although it would be interesting for future work to identify if there are autism specific visual experiences, including individuals with these co-occurring conditions, due to their high prevalence in autism, is relevant for providing a realistic description (Gillberg and Billstedt, 2000).

CONCLUSION

This study provides a first-hand insight into the range of visual issues and their impacts within the autistic adult community which cannot be expressed through objective or quantitative studies. The findings have confirmed that autistic adults are often dissatisfied with their vision and experience a range of visual sensory symptoms which vary from person to person. These symptoms can occur alone or as part of a larger multisensory response, nevertheless, vision contributes to sensory issues and emotional responses. It is noteworthy that although some of the visual experiences expressed by our participants can be expected to occur in non-autistic people, it was the magnitude, frequency and impact of these experiences which was unique and suggested to be greater. Although a large part of the visual experiences suggest issues with higher level processing, there is indication that some symptoms associated with control of binocular vision and visual stress could benefit from an optometric assessment. Finally, autistic adults employ a variety of strategies to overcome their visual symptoms, but the last resort is to endure these.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by The University of Manchester's Research Ethics Committee (UREC). The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

KP, EG, CD, and CP led the study, designed the study, recruited the participants, conducted the data analysis, and wrote the manuscript all with the support of his supervisors. EG, CD, and CP were involved in designing the study and finalized the data analysis themes. EG and CD supported the focus group sessions

which were facilitated by KP. JP and PB were advisors for the duration of this study and gave suggestions to ensure the study was accessible to autistic adults. All authors were involved in editing the manuscript and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.633037/full#supplementary-material

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"No Way Out Except From External Intervention": First-Hand Accounts of Autistic Inertia

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This study, called for by autistic people and led by an autistic researcher, is the first to explore 'autistic inertia,' a widespread and often debilitating difficulty acting on their intentions. Previous research has considered initiation only in the context of social interaction or experimental conditions. This study is unique in considering difficulty initiating tasks of any type in real life settings, and by gathering qualitative data directly from autistic people. Four face-to-face and 2 online (text) focus groups were conducted with 32 autistic adults (19 female, 8 male, and 5 other), aged 23–64 who were able to express their internal experiences in words. They articulate in detail the actions they have difficulty with, what makes it easier or harder to act, and the impact on their lives. Thematic analysis of the transcripts found four overarching themes: descriptions of inertia, scaffolding to support action, the influence of wellbeing, and the impact on day-to-day activities. Participants described difficulty starting, stopping and changing activities that was not within their conscious control. While difficulty with planning was common, a subset of participants described a profound impairment in initiating even simple actions more suggestive of a movement disorder. Prompting and compatible activity in the environment promoted action, while mental health difficulties and stress exacerbated difficulties. Inertia had pervasive effects on participants' day-to-day activities and wellbeing. This overdue research opens the door to many areas of further investigation to better understand autistic inertia and effective support strategies.

Keywords: autism, movement, inertia, catatonia, qualitative, autistic adults, ASD, initiation

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INTRODUCTION

Autism is a heterogeneous condition viewed as primarily a disorder of social interaction accompanied by rigid and repetitive thinking and behavior. Sensory and motor differences are mentioned only peripherally in the diagnostic criteria (American Psychiatric Association, 2013; World Health Organisation, 2020); however, sporadic but increasing research over the last 25 years has proposed that these differences may be more important in the etiology than commonly thought (Leary and Hill, 1996; Robledo et al., 2012; Donnellan et al., 2013; Torres and Donnellan, 2015; Breen and Hare, 2017; Welch et al., 2020). They have proposed rethinking some autistic

traits, and associated behavioral issues such as non-compliance, as manifestations of sensory and motor differences.

An experience commonly known in the autistic community as 'autistic inertia' may be attributable in part to differences in motor control. 'Inertia' is the term for Newton's first law of motion, which is the tendency of a body to stay in the same state of motion unless acted on by an external force. This is used metaphorically to describe difficulties both starting and stopping activities, which are commonly experienced by autistic people[1]. Inertia is described in personal blogs (Paterson, 2016; Sparrow, 2016; Buckle, 2017; Welch et al., 2020) and discussed in autistic groups and events. Inertia overlaps with the concept of 'monotropism' (Murray et al., 2005), or the autistic tendency to focus narrowly and deeply on topics or objects of interest, which has both positive and negative aspects. Difficulty with initiating action, specifically, is usually experienced as problematic. In response to an article on monotropism, an autistic author writes:

> *To me it seems odd that inertia is often so far down the list of things that people associate with autism? [. . .] I find [inertia] probably the single biggest problem I have that stems directly from it.* (Murray, 2017).

As evidenced by the first author's lived experience as an autistic person who experiences severe difficulties of this nature and witnesses them in others, autistic inertia, and its effect on executing intentions, is a crucial topic to address. Although there is widespread recognition of inertia within the autistic community, and the significant effect it can have on autistic people's daily lives, no formal research has directly investigated its nature or impact. Increased awareness and understanding of initiation impairments is particularly important as the ability to spontaneously initiate voluntary actions may underlie or influence some social and behavioral differences that are characteristic of autism.

Although there is some indication that motor difficulties are a factor, the term 'autistic inertia' may be an umbrella term for impairments with distinct aetiologies. For example, in her blog, Tanea Paterson, an autistic adult, describes inertia in terms of difficulty initiating movements, following instructions, and flexibly changing attentional focus (Paterson, 2016). Others describe inertia as an inability to act due to fear of unknown or undesirable outcomes. It is unclear from the existing literature whether these difficulties with initiation arise from: (i) social-emotional factors such as a primary social impairment or mental health difficulties (Hollocks et al., 2010), (ii) executive dysfunction (Ozonoff and Jensen, 1999), (iii) movement differences such as catatonia (Wing and Shah, 2006; Breen and Hare, 2017), or another mechanism as yet unidentified. These possible mechanisms are underpinned by overlapping neural circuitry, which have been found to function differently in autistic people (Abbott et al., 2018; Ozsivadjian et al., 2020).

Nearly all research on initiation in autism is intervention-based research with autistic children using the frequency of social initiation as an outcome measure. Researchers consider various mechanisms for diminished social interaction in autistic children, such as lack of social motivation (Chevallier et al., 2012; Kohls et al., 2012) or learned helplessness (Koegel and Mentis, 1985); however, none consider the possibility of an underlying deficit in the ability to initiate actions.

Mental health difficulties such as depression and anxiety occur at high rates in autistic people (Hollocks et al., 2010; Hudson et al., 2018). Avoidance due to anxiety or lack of motivation due to depression both could contribute to a lack of initiative. These issues have been linked to difficulty understanding and processing one's emotions (alexithymia), intolerance of uncertainty (Neil et al., 2016), and cognitive inflexibility (Ozsivadjian et al., 2020). Anxiety can lead to avoidance, and depression to loss of motivation. Because of the internal, subjective nature of motivation and initiative, it is difficult to distinguish between emotional and other drivers for failure to act.

Alternatively, initiation difficulties could be due to impairments in executive function (skills involved in planning, working memory, attention, and inhibition), which have consistently been found to be impaired in autism (Ozonoff and Jensen, 1999; Bramham et al., 2009; Brandimonte et al., 2011; Demetriou et al., 2018). Monotropism is a framing of autistic attention distribution as a tendency to narrow, intense focus that contributes to autistic strengths such as expertise and enhanced detail perception (Murray et al., 2005). When viewed as a deficit, this fixed focus is known as 'cognitive inflexibility,' an aspect of executive dysfunction associated with anxiety and depression (Ozsivadjian et al., 2020). Differences in the cortico-striatal circuitry underlying these functions has consistently been found in autistic children and adolescents (Abbott et al., 2018; Uddin, 2021). Autistic people are also found to have impaired prospective memory, i.e., remembering to do something later. Providing a cue or initial step has been found to reduce initiation-specific deficits (Williams et al., 2014; Carmo et al., 2017). Social interaction may be particularly vulnerable to the effects of initiation impairments because it is variable and unpredictable, calling on a variety of high level flexible cognitive processes (Riggs et al., 2006).

Finally, difficulties with initiation may stem from a movement impairment. Unusual patterns of movement have been observed in autism since its first descriptions, and motor coordination difficulties have been found in up to 80% of autistic individuals (Fournier et al., 2010; Gowen and Hamilton, 2013), yet, with the exception of repetitive movements, motor symptoms are usually treated as being peripheral or additional to autism (Leary and Hill, 1996; Ming et al., 2007). 'Autistic behaviors' such as non-compliance, lack of communication, lack of affect and resistance to change could be due to difficulties with initiation of movement. Ming et al. (2004) reported this phenomenon in a single case of an adolescent autistic girl who was considered severely non-compliant. The participant was instructed to squeeze a hand grip while muscle action and physiological correlates of mental effort were measured. Indications of internal effort were seen even when no muscle action was recorded. Further evidence of a mismatch between intentions and actions comes from analyses

[1]This paper uses the term 'autistic people' to reflect the preferences of a majority of autistic adults (Kenny et al., 2016), although the authors acknowledge that this preference is not universal, with some finding it offensive (Bury et al., 2020) and that research on the topic remains scarce (Shakes and Cashin, 2019).

of the memoirs of three minimally verbal autistic young people (Welch et al., 2018) as well as the writings of autistic bloggers (Welch et al., 2020). Both of these papers included several themes related to initiation problems, e.g., 'I can't start my body' and 'Brain-body disconnect.'

Slow movement and delayed initiation have been reported in autism since the mid 1990s (Leary and Hill, 1996). This was first detailed in the context of catatonia in 2000 (Wing and Shah, 2000). Catatonia, a complex psychomotor syndrome, is typically envisaged as a lack of responsiveness to the environment (stupor) and freezing in awkward positions (posturing); however, Wing and Shah described a range of difficulties including extreme slowness, freezing mid-movement, prompt dependence, repetitive movements, mutism, and deterioration in self-help skills. The full catatonia syndrome occurs in up to 20% of autistic people (Wing and Shah, 2000; Billstedt et al., 2005) and nearly half of a group of 87 autistic adolescents were found to have clinically significant catatonic features (Breen and Hare, 2017). As with cognitive flexibility and repetitive behavior discussed previously, catatonia may be associated with abnormalities in thalamocortical loops although the exact mechanisms have yet to be clarified and there are indications of diffuse pathway dysregulation (Daniels, 2009). Wing and Shah expanded on their description of catatonia in autism in a paper exploring catatonia-like features without marked deterioration in autistic children and adults (Wing and Shah, 2006) and numerous cases are detailed in Shah's recent book on the subject (Shah, 2019). In the literature, the most extreme forms of these motor issues are usually associated with 'severe' autism, it is now understood that catatonia can have a range of expression from the most recognizably severe manifestation to mild and intermittent. It is possible, therefore, that more subtle expressions of catatonia are under-reported within the autistic spectrum more widely.

Despite the high prevalence of catatonia-related phenomena and the severe impact on functioning, autistic catatonia has been under-explored in research. Moreover, due to the severe disability of those who have been studied, what research exists has been based almost exclusively on second-hand reports from carers and clinicians. However, milder expressions that would not be readily recognized as catatonia may share some underlying characteristics. There are obvious limitations to the understanding that can be gained by observation of a condition characterized by impairments in action and expression. Welch et al. (2020) explored autistic embodiment through analysis of blog posts by both speaking and non-speaking autistic people. They found some difficulties regulating movement which were reminiscent of catatonia-like impairments. While this work contributed to the understanding of internal autistic experience, those who experience the most significant impairments in voluntary action are unlikely to be able to consistently write a blog. The present study further explores these issues, focussing specifically on the ability to act on intentions, by talking to autistic people who share some of these difficulties. In particular, characteristics that are often invisible to observers such as difficulty initiating, emotional states, and motivational factors may be clarified by first-hand reports.

In summary, inertia is commonly reported by autistic individuals, but has not previously been the focus of any formal research. In order to explore the nature, mechanisms and impact of inertia, this study used first-hand descriptions, collected via focus groups, of difficulties autistic people experience with doing things they need or want to do and their impact on day-to-day life. This is an important topic for the autistic community, with implications for our understanding of and approach to a subset of autistic behavior that creates challenges both for caregivers and for autistic people themselves. The lead author's personal interest in the subject inspired her to attempt to assist autistic people suffering from these difficulties on an individual basis as well as organizing informal discussion groups on 'autistic inertia' and 'catatonia' at Autscape (an annual residential event for autistic people) in 2017 and 2018, respectively. Each of these groups was attended by approximately 40 autistic individuals who shared their experiences and provided mutual support. Participants in these groups strongly advocated for research into the subject, and these discussions have informed this research.

MATERIALS AND METHODS

Approach

The aims of this research were to explore the experience of inertia and to begin to describe these experiences and their impact. So little is known about this collection of difficulties that a broad approach was required and a realist framework was adopted for this initial description. Following the success of previous discussion groups at Autscape, which gave participants an opportunity to share their experiences and hear those of others, data was collected through focus groups, both face-to-face and online. Previous research on related topics has used only observation, second-hand reports from carers and writings of autistic people, so first-hand reports allowed for unique insights into the internal experience of autistic inertia. This study was approved by the University of Manchester Research Ethics Committee (ref. 2019-6324-11577).

Recruitment

The face-to-face focus groups took place at Autscape, a well-established annual residential event in the United Kingdom organized by and for autistic people. Autscape does not allow researchers to approach potential participants to avoid pressure or coercion, so recruitment was entirely by the placement of posters and sign-up sheets for the group sessions. The sample was purposive; the recruitment posters referred to experiences of getting stuck or having difficulty doing things. Despite the restrictions on recruitment methods, two additional groups were needed to accommodate the high number of volunteers.

Two further online (Skype) focus group sessions were conducted because many autistic people have difficulty with travel to unfamiliar places and interaction in groups. Text rather than video chat was used in order to maximize access because autistic people often have difficulty with various aspects of social communication such as the timing of conversation turns, auditory processing, and attention. Several adjustments were

needed to improve accessibility to autistic people, for instance by requesting that moving images not be used to reduce the visual processing stress. More detail about conducting text-based meetings with autistic participants can be found on the website of autism research charity, Autistica (Buckle, 2020) and the Autism@Manchester website (Buckle and Gowen, 2021).

In order to obtain the widest representation possible, the selection criteria were kept to a minimum, with no exclusions for psychiatric or other conditions which commonly occur with autism. The requirements were that participants must be age 18 or over, clinically diagnosed with any autism spectrum disorder and able to express their experiences in words.

Participants

Informed consent was obtained from all participants prior to engagement in the study. Demographic data were obtained with a brief written questionnaire. Participants consisted of 32 adults age 23–64 years (mean = 45). Their self-described genders were: 19 female, 8 male, 4 non-binary, and 1 unspecified[2]. All had a clinical diagnosis of any autism spectrum condition by a suitably qualified clinician or multi-disciplinary team. Where possible ($n = 14$), diagnosis was verified by having sight of the participant's diagnosis letter. Where the diagnosis letter could not be obtained, details of the diagnosing clinician, clinic and date were taken.

The remaining background questions, which were not answered by three participants, are reported here in order to fully characterize the sample. All but one participant (who lived with their parents) lived independently in the community. Half of the remainder ($n = 14$) lived alone. Of those who lived with others: 4 with a partner, 3 with their children, 6 with partner and children, and one with a flatmate. 18 individuals received care or support due to their disability. Nine of those who responded worked full time, 7 worked part time, 4 were students, and 9 were unemployed or retired. Participants reported the following mental health diagnoses: 24 had anxiety, 20 had depression, 12 had been diagnosed with PTSD (5 had recovered), 2 had a past diagnosis of psychosis, but one considered this to be a mis-diagnosis before their autism was recognized. Eight reported that they were taking neuroleptic medications or had in the past. They also reported the following neurological conditions: 8 ADHD, 7 dyspraxia, 8 migraine (not asked on form) and one had a diagnosis of catatonia. Additionally, 4 reported a fatigue-related condition such as chronic fatigue syndrome.

Procedure

Six focus groups were held, each attended by 4–6 participants. Details of the composition of each group are given in **Table 1**.

Four face-to-face groups were held over the 3 days of the Autscape event in July 2019, and a further two groups were held online in May 2020. Online focus groups were conducted after the data from the face-to-face groups had been collected and analyzed. The lead author (KB) conducted all 6 groups, and participants were made aware that she is also autistic and experiences significant difficulty with initiation. In addition, one online focus group was attended by KL and the other by EG. Each meeting lasted 1.5–2 h. The initial half hour provided an opportunity for participants to familiarize themselves with the research and procedure for the session and to ask any questions about what would happen. Participants were encouraged to talk about their experiences of difficulty doing things they want or need to do. Questions were oriented around 'difficulty doing things' because the researchers anticipated from background understanding that these may not be easily segregated into difficulty initiating a task vs. difficulty stopping an ongoing activity in order to initiate a new one. Questions were asked according to a schedule to prompt a range of responses (**Table 2**). Any participant who had not contributed was specifically invited to do so before moving on to the next question, with an explicit option to pass. This was rarely needed and nearly all participants responded to all questions. Face-to-face groups were audio recorded and later transcribed by a professional transcription service.

Analysis

All face-to-face interviews were completed prior to transcription due to time constraints at the Autscape event. Text transcripts of the audio recordings were carefully checked against the recordings for errors or omissions. The text from the online groups was used as written by the participants with minor corrections of punctuation and spelling. Data about gender, age, support needs and co-occurring conditions were collected in order to fully characterize the sample, and to indicate possible avenues for further research. Additional diagnoses were not verified. Therefore, in this study, data were not analyzed separately according to additional conditions.

Data analysis was conducted using inductive thematic analysis, following the reflexive method set out by Braun and Clarke (2006, 2019, 2020). Because the aims were concrete and descriptive, a realist framework was used in which the experiences of the participants were coded and interpreted on a semantic level, without reference to social context or unarticulated meaning. After familiarizing herself with the data, KB exhaustively applied codes to each concept present within the data. The codes and categories were reviewed, analyzed, refined, categorized, and combined to generate themes and group them in meaningful categories. This was an iterative process and developing the structure to include a manageable number of themes required reanalyzing the codes, categories and themes several times. Saturation was reached after five groups, with no new themes arising in the final (sixth) group. Coding was conducted only by KB, who then discussed and refined themes in collaboration with all authors, with a selection of participants (described below) and again following peer review.

KB made an effort to reflect on her lived experience and prior exploration of autistic inertia, and the influence this would have in analysis. In particular, KB had a pre-existing belief that autistic inertia cannot be explained by anxiety alone and there is a movement component to initiation difficulties experienced by some autistic people. KB also has influence as a leader within the Autscape organization which may have affected participant

[2]In recognition of the fact that alternative gender identity and expression is common among autistic people (George and Stokes, 2018), the questionnaire asked all participants to self-describe their gender rather than providing specific options. Those who do not identify as either 'male' or 'female' often refer to themselves as 'non-binary'. At times this paper will use the singular 'they' to reflect the preference of some participants to avoid using gender-specific pronouns.

TABLE 1 | Focus group composition.

Group	Format	*n*	Age mean (range)	Gender		
				Male (*n* = 8)	Female (*n* = 19)	Other (*n* = 5)
1	Face-to-face	4	46 (32–64)	2	2	0
2	Face-to-face	6	49 (37–62)	0	4	2
3	Face-to-face	5	43 (33–53)	2	2	1
4	Face-to-face	6	45 (36–51)	3	3	0
5	Online	6	44 (25–58)	1	4	1
6	Online	5	45 (23–45)	0	4	1

Other includes both non-binary and unspecified gender.

TABLE 2 | Focus group questions and prompts.

What are some experiences of difficulty doing things?

Prompts:

- Do you have any specific examples of when you've been unable to do something you needed or wanted to do?[a]
- Are they things you want and are motivated to do?[b]

What makes it harder?

- What do you think stops you from getting things done?

Prompts:

- Do you get paralyzed with anxiety?[b]

What makes it easier?

Prompts:

- Plans, schedules, or alarms[b]
- Someone else starting it off[b]
- Does music have any effect?[a]

Can you describe what it feels like to be 'stuck'?

Prompts:

- Do you feel like you've slowed down (or everyone else has speeded up)?[b]
- Do you feel anxious?[b]
- Do you know how much time is passing?[b]

Does this have an impact on your life?

Prompts:

- For example, your ability to be productive – study, work, parent, volunteer, etc.[a]
- Your ability to take care of yourself[b]

The main questions (in bold text) were asked of each group. Prompts were only used if no one in the group spontaneously mentioned the topic.
[a]Prompts that were used in most or all groups.
[b]Prompts that were used in a minority of groups.

responses; however, this background also contributed to trust and rapport within the groups. A visual record of the development of the themes was maintained in order to review decisions and confirm that important concepts had not been lost.

Validation

A selection of participants were consulted throughout the analytic process, which helped to shape the themes and final structure. The results were presented to many of the participants and others following Autscape 2020. Participants confirmed that the analysis was an accurate representation of their experiences. One responded that the experiences described were so close to their own that they could not readily identify which quotations were theirs. Another said that reading about others' experiences helped him to be more forgiving of his own difficulties. Participants approved of the theme structure and analysis without any requests for corrections.

RESULTS

The present study investigates autistic people's experiences of difficulty doing things they need or want to do. Topics arising during focus groups that were not related to this (e.g., general attitudes about autism and experiences of the Autscape event) are not included. The autistic community jargon of 'inertia' was often used to refer to difficulty stopping, starting and changing tasks. A diagram of the themes is provided in **Figure 1**. Participants related their experiences objectively and analytically, with honesty and candor. They provided detailed descriptions of their difficulties in considerable depth, and these fell into two main categories: descriptions of inertia itself, and its effects. Within each of these categories, four themes reflected internal experiences and two themes related to how inertia interacted with the external world. Each of the themes is described with illustrative quotations from the data. Quotations are provided verbatim and names are pseudonymised to protect privacy.

Descriptions of the Internal Experience of Inertia

Four themes described core characteristics of participants' internal experiences of inertia: tendency to maintain one state, lack of voluntary control, difficulty finding the first step and disconnection between intention and action.

Tendency to Maintain One State

Participants were asked about problems 'doing things'; however, in their responses, difficulty acting encompassed not only starting, but also stopping and switching tasks.

> *I can't get to the point where I'll go to do the thing because it's almost like I got to stop whatever I'm doing, whether I'm doing anything or not. Even stopping not doing anything is stopping doing something. – Ruth.*

Because of this difficulty differentiating starting from 'stopping not doing anything,' and true to the 'inertia' metaphor, the term

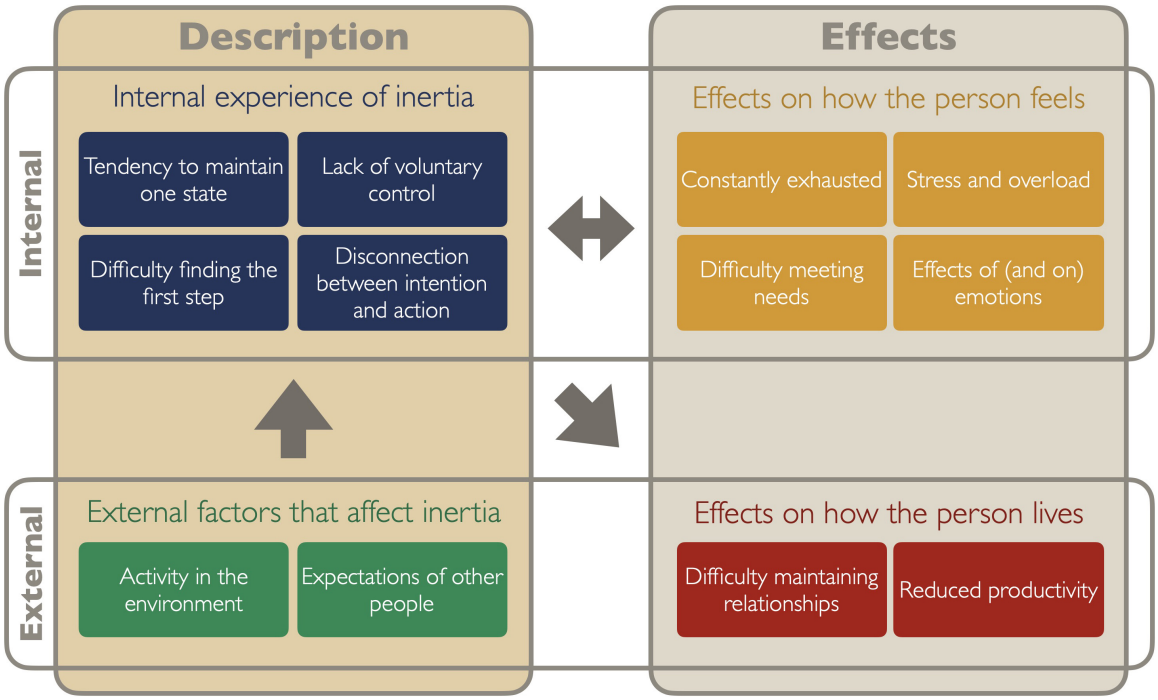

FIGURE 1 | Diagram of themes in autistic inertia. Themes (colored rectangles) are organized into primary categories (columns) of 'Description' and 'Effects.' These are further divided into internal and external domains. This then provides four sub-categories: internal experiences of inertia (blue), external factors that inhibit or facilitate action (green), effects on how the person feels (yellow), and effects on how the person lives (red). Arrows indicate the direction of effects as one area influences another.

'inertia' is used to refer collectively to these related functions, especially when these interfere with initiation of new activities.

Continuity of a task made it easy to stay in the same state of activity or inactivity; interruptions would disrupt that, which could be helpful or harmful depending on whether the person wanted to switch. Several participants stated that starting was more problematic than sustaining action, e.g., Ruth said, *"I'm alright once I get going but getting going can take a long time."* As an indirect consequence of difficulty stopping, they would sometimes avoid engaging in certain activities for fear of being unable to break away when they wanted to.

Once inertia was 'in motion,' any disruption could derail a task completely. Participants were unable to suspend a task for an interruption and pick it up again, which at times made them reluctant to even begin. Similarly, having to interrupt a task to do something else, such as to fetch a necessary item, would then make it very difficult to re-start the task.

> *I'm in the office and there's paper everywhere and I'm trying to sort it out. And then I do have to go and do something else, so, you know, the child needs to eat food or whatever. Like I just can't get back to the place I was before. So, I can't get back to the task. And then it's even worse than it was before. The fear of that happening stops me from starting a new task. – Elizabeth.*

A break in continuity, such as any kind of barrier, could make a task impossible to initiate or sustain. The barrier could be (a) physical, such as an item blocking access to the washing machine; (b) social, such as having to walk in front of another person; or (c) psychological, such as having to make a decision:

> *I spend the whole day not quite deciding whether to have the shower first or do something else first or do a load of laundry, and then maybe go out after doing the laundry or go and get it over with. – Daniel.*

Perfectionism and a desire to make the ideal choice exacerbated these issues.

Conversely, strategically-placed barriers and visual cues could interrupt a habitual action and make it easier to engage in a desired activity. For example, placing an instrument and music in the way of getting from one room to another. At a more basic level, an interruption could trigger action for someone who was frozen.

> *Sometimes, I end up just sitting and not doing anything when I really want to be reading a book that's right next to me, but I'm not. And that can last often till there's an external sort of interruption, which is normally like if my partner walks past the door, then I'll be like, 'Oh, I should move now.' – Lisa.*

Difficulty starting occurred most often with procedural barriers, such as having to put on shoes, or where a sequence of actions was needed:

> *There are always things that need to be done, before I can do 'the thing' – so, I want to brush the hall floor (not sure why) but first I need to move things from hall floor. Move crutches. . . where to?*

Somewhere accessible, but not in the way – sidetracked. scissors picked up – oh, now I've found scissors I can. . . Sewing machine still there after 5 months – need to put it away, but there's no space for it and my son tidied my shed, so must keep it tidy. Exhausted, lie down! – Harriet.

Such barriers typically not only blocked the task they were trying to do, but also transitions to another task. The bigger the barrier, the more difficult it was to start; more significant changes of position (e.g., getting up) or transitions (e.g., leaving the room or house) were more problematic than starting tasks that didn't require such transitions.

It takes me an awful lot to get out of the house. There's so much that has to be done to get out of the house at vaguely the right time with all the stuff that you need and properly dressed and so on. And I actually find if I can put all that preparation in – which is a lot of preparation – for all the things today and go out and not come back home again so it's done and go from one activity to another activity to another activity. It actually takes a lot of the pressure off because the hardest thing for me of all is leaving the house. – Nicky.

Strategies used to overcome these issues often involved reducing barriers, lowering the threshold to action. Several participants talked about getting themselves to start something by consciously breaking it into small steps and talking themselves through it. For example, when having difficulty getting out of bed:

Eventually saying to myself, 'All you need to do is. . ..' And so, all you need to do is sit up, sit on the bed. All you need to do is sit on the edge of the bed, stand up, you know, walk out of the bedroom to the bath, et cetera. And then I suppose similarly getting dressed – Fred.

Sometimes they would deliberately avoid stopping an activity because they feared being distracted or becoming stuck in an inactive state, and unable to return to their original activity.

I find sometimes I have to say to myself 'Don't sit down, don't sit down, don't sit down yet. Okay, now you can sit down.' – Emma.

Although the focus of the discussion was on the difficulties of not being able to do things they need or want to do, the tendency to keep going could help to get tasks completed or was desirable in itself. These were to do with the ability to, as Paul said, *"become totally immersed in some things."* This could be specific to the task at hand, or a more general feeling:

I'd like to mention the flipside to all of this when everything goes right. I do everything with extreme capability, and everything is just right, and that other thing that's nagging, pestering at the back of my mind is not present and it's like it takes no time. Everything feels like it's so fast and you do everything so quickly. – Joel.

Deep immersion in a desired activity was described as pleasurable, even if it involved losing track of time, again as Joel contributed, *"When I'm focused on something, I'm not aware of how much time's passed. It could be an hour. It could be 6 h."*

Lack of Voluntary Control

The other central aspect of their difficulty was that it was experienced as involuntary and impervious to their conscious efforts. For example, Margaret wrote: *"I also can't overcome my inertia. I have to wait for it to go away."* Despite the insightful and detailed descriptions of the problem, many felt that their problems were unpredictable and largely incomprehensible, describing them as 'ridiculous' and 'illogical.' In particular, they were confused by their inability to execute tasks that were within their capability, and by the variability of both the expression of the problem and the effectiveness of strategies.

Yes, I never manage to schedule cooking for the week, but often when I do manage to cook I make several meals worth. It's just I can't guarantee to manage that when I need to. – Naomi.

This effect was quite noticeable in the difference between an established routine and spontaneous action.

Routines. Routines help a lot. Anything that I can routinise so that I don't have to think about it quite so hard helps. And then I can do those things a bit more on autopilot. – Lisa.

Conversely, a plan that required internal effort to execute could be impossible, despite sometimes acute awareness of the consequences:

I think it's the expectation as well. Like for example, I know I can't switch between tasks during the week, so I decide to cook all my meals on Sunday and meal plan. But then Sunday comes and the pressure of doing this thing in that specific timeframe is really too much, so I struggle to do it because of that. Instead, I spent the day panicking and being mad at myself for not doing it and thinking of how much it's going to ruin the rest of my week. – Margaret.

They reported having to manipulate themselves into doing things.

I just don't feel like I have control over what I'm doing necessarily. I feel like I'm coaxing myself through things or I'm trying to work out strategies to make myself do things. – Alex.

They often tried to convince themselves to act, but this was usually ineffective:

My head is saying all the right things, like you'll feel better if you do X, or if you get up now, you'll be able to do that thing you're excited to do, but it's like the rest of me is a stubborn child. . . I think there's some demand avoidance in there too. – Jo.

Together, the tendency to stay in the same state and the lack of voluntary control were threads that ran through descriptions of inertia and the factors that influenced it.

Difficulty Finding the First Step

Several participants felt there were different issues underpinning their difficulty acting, for example:

It's almost like there's two different kinds of things. It's kind of like there's the kind of the mental kind of plan-y sort of stuff, which is more kind of stress-inducing almost. And then there's a different kind [. . .] almost like a physical thing where you just kind of get physically stuck. – Ruth.

Difficulties with planning, and with executing a plan, were a major issue. Some had difficulty breaking down a task or formulating a plan:

Too many different things need doing – can't prioritize. Very easily overwhelmed by amount/number of things to do. [. . .] Difficult to separate the 'blob' of 'lots to do' into small, potentially doable bits. - Harriet.

Some had difficulties that were almost the opposite. They could break down a task, but continued to break it down until it had so many competing elements they were unable to see how to proceed:

I can cook. But a lot of the time, I buy the ingredients and I never cook anything because it gets too complicated in my mind. – Joel.

Others expressed difficulties to do with prioritizing or finding a starting point. They often needed the help of another person to work out how to approach a task.

It's having a tornado of things going through your head, trying to work out how to focus on one thing and work out how to pick one thing. Some might be tasks to do (fun or not), some will be processing the day or specific info, all of which makes it harder to find a way in to the 'to do' things. - Jackie.

A weak working memory could create issues with planning and with executing any plan. Alex relates it like this, *"I will go 'oh, I'm in this room now, what was I doing? I was doing this thing, I'll go and do that – oh no, apparently, I've done that already."* Alex could forget whether they'd used their toothbrush, whether they'd taken a puff of inhaler seconds earlier, or even whether they had already made a decision.

Disconnection Between Intentions and Actions

In addition to planning difficulties, a subset of participants also felt, at times, that there was a disconnection between their perception, emotions, intentions and actions. Unlike the difficulties related to planning and prioritizing, which tended to affect complex tasks, disconnection between intentions and actions could apply to actions that seemed simple. Thomas said that, *"It seems ridiculous sometimes. You just can't do certain things that seem so simple,"* and Lisa provided an example:

For some things like I find it really difficult to work out why I'm not getting started especially when it's something I really want to do. [. . .] and there are only one or two steps for me to start doing it like picking up a book that's right next to me. I just don't. . . I don't understand why that's so difficult sometimes.

This experience of disconnection had three distinctive characteristics: feeling physically unable to move, altered awareness, and passivity.

Physically unable to move

Participants described their experience as 'physical' and that although they knew what to do, they 'just can't.' Although analysis was not conducted with *a priori* codes, it was recognized that this description had several characteristics in common with catatonia, although sometimes with a more subtle expression. Examples of these are provided in **Table 3**.

Some participants found themselves unable to take a specific deliberate goal-directed action while still being able to move in other ways. For example, Daniel would struggle to get out of bed, despite being thirsty and needing to use the toilet, even while he could play with his phone.

Sometimes, a drink is actually. . . maybe not in arm's reach, but in like standing up a little bit and reaching, reach. I can't understand why I won't get it. And afterward, I think. . . how was that about? Why did you give yourself a headache and do that for 45 min or something until you're almost on the verge of wetting your bed? And then, what's going on? Am I myself when I'm doing that? Is somewhere the physiological thing taking over? What is the problem? . . . It's not an every day thing for me, but [. . .] it usually

TABLE 3 | Examples of catatonia-like features from participant reports.

Catatonia	Example of related experience from focus groups
Periods of shutdown[a], being very still for long periods of time[b]	*I'm going to make a drink and standing and then realizing an hour later that I'm still standing in front of the kettle and haven't actually done anything. I'm thinking but why, how has that happened? - Sam*
Movement difficulties (freezing and getting stuck)[a], getting 'stuck' when trying to complete actions[b]	*It feels like I'm holding my breath and my body is frozen. It's a literal inertia. I mean it's a literal paralysis and very often I will find that I am actually holding my breath. . . So the feeling of it is literal, nothing moving, no thought, no breath, no movement. - Emma*
Difficulty stopping actions once they have been started [b]	*I find one of my things is reading news websites now, and that I just end up reading sometimes different articles and sometimes sort of the same one over and over. - Lisa*
Difficulty initiating actions[b]	*I'm finding it really difficult to actually just pick up a book and get started. - Lisa*
Increased slowness[a], moving very slowly[b]	*I feel I can move but like really slowly and only to like lie down or curl into a ball as I feel frozen or freezing up. - Kelly*
Prompt dependence[a], taking a long time to finish actions or requiring prompts to complete actions[b]	*If I'm struggling to get to bed and my partner has already gone to sleep, chances are at some point, he might get up to go to the loo or at least I know at some point he's going to get up in the morning. And that will probably unstick me. . . - Lisa*
Movement abnormalities[a]	*When I put the washing machine on, I find I spend an hour in the kitchen [. . .] kind of swaying around or juggling or just doing things [. . .] And somehow, that washing machine, when it's on it's like I'm magnetized into the kitchen. - Daniel*
Passivity and apparent lack of motivation[a]	*If I'm wanting to do a social thing, I'd like to spend time with people, I have difficulty initiating [. . .] I wouldn't even sometimes think to contact them to start it. I'll just go, I would have liked to have been doing something with somebody and it's been quite a new thing to realize that I can start a conversation with somebody like somebody else doesn't have to start the conversation first which isn't always evident. - Ruth*
Posturing[a]	*It's like im frozen in time. . . at worst it can hurt cause it feels like I want to move but can't - Brian*
Fluctuations of difficulty[a]	*I just find myself utterly baffled when I'm just stuck. And sometimes, I'm stuck for days on end and just in the contrast with how productive I can be on other days. It just baffles me. – Elizabeth*
Catatonic excitement[a]	Not evident in the data.

Characteristics of autistic catatonia drawn from two sources, as indicated: [a] *Primary difficulties and manifestations of autistic catatonia (Shah, 2019, p. 29).* [b] *'Core' features of catatonia in autism from Attenuated Behaviour Questionnaire (Breen, 2014).*

eventually results in pain, dehydration headaches, things like that. – Daniel.

Altered awareness

In addition to describing physical difficulty moving, participants talked about their internal experience during such episodes. Sometimes the person felt disconnected from their physical body, thus unable to control it.

Sometimes when I feel stuck, if it's. . . where I feel I can't for example get off the sofa, it's almost that dreamlike state where I can hear everything but it feels kind of slightly far off. – Suzanne.

Sometimes they felt 'stuck in their mind,' unable to enact things with their body:

I can't unpick what I need to do, where to start or how to find the energy to get beyond the thinking about things. . . and then 3 h just goes out the window - Brian.

This could be combined with altered time perception, for example:

I am aware of my surroundings, but time feels slower, more drawn out and I don't remember being able to feel my body other than being frozen but it feels as if I go completely into my head, like an out of body experience but in my mind. – Kelly.

At times, they could even experience a complete cessation of thought, awareness and action, so the person would find themselves in exactly the same position, but between several minutes and several hours had passed.

Sometimes, I'd be like, 'Oh, I want to read. Oh, here's a book.' And then I'm reading the book. And sometimes, I'd be like, 'Oh, I want to read.' And then it's 3 h later and I haven't moved. – Lisa.

Passivity

Even while conscious of the inability to perform a desired action, there was minimal sense of physical or mental strain. They seemed to simply accept that the desired movement did not happen.

I'll be sitting on the bed thinking I should really go to bed. I really want to go to bed. I'm really tired. But it's just not happening, but it doesn't worry me, the way that sometimes things really worry me. – Lisa.

Like Lisa, others often had little emotional arousal about the situation.

It feels like what I'm thinking is sort of somewhere out here, kind of passively observing myself and going, 'hmm. I'm not actually doing that thing that I want to do. I'm not sending the text message I could be texting. I'd quite like to contact the person.' So, I'm not like stressed about it or anxious. . . . it's like commentating on it but in very sort of, 'oh, that's mildly interesting' sort of way. – Erin.

Descriptions of External Factors That Affect Inertia

In the absence of internal drive, participants found themselves dependent on the scaffolding provided by external cues and prompts.

Like I am stranded in the middle of the sea and nothing exists anymore. There is no past, no present, nothing to do and no way out except from external intervention. – Margaret.

The key external factors, activity in the environment and expectations of other people, could both facilitate and inhibit action.

Activity in the Environment

Several participants described human and non-human elements of their environment prompting and sustaining action. Environmental cues, like an office environment, and synchronous activity, such as someone working on a similar task nearby, could help the autistic person to do the same.

Sometimes, what helps me is having another person present, but I don't necessarily want them to interact with me. Just there working beside me, maybe doing the task with me, but not. . . just working side by side just kind of motivates me for some reason. – Daniel.

Conversely, asynchronous activity or irrelevant movement and background noise was usually distracting and stressful. This was most pronounced in the highly varied responses to music. Some found that music put them in the mood to act and could make it easier, while others found it a problematic distraction.

Expectations of Other People

The most often reported helpful factor was the assistance of another person; however, the influence of others could also hinder action if it was stressful or demanding. Several participants said that prompting by another person could be very helpful for getting unstuck.

The only thing that helps me, only thing that works, and it works consistently, is just to have a stuck buddy that I text. . . . And all I have to do is text, 'I'm stuck.' [. . .] And we just text it out and kind of make a plan. – Elizabeth.

Being expected to do something for or with someone, such as by scheduling an activity with another person or being needed, was often helpful:

It's much easier to do something for another. I can even do form filling with another person and I'm hopeless with forms. So for somebody else then yeah, that makes me do it. – Nicky.

The most effective supports were time sensitive. A sense of urgency could make even stalled actions possible for some participants:

Sometimes having to do something straight away helps. Once a friend asked for a cake recipe but kept saying no pressure, when you're ready, and I failed and failed to send it for weeks, Then 1 day she emailed and said she needed it for tomorrow when she had visitors coming and I just did it straight away! After all those weeks. – Naomi.

Some participants recognized this and deliberately scheduled external time-sensitive activities such as having to be somewhere at a specific time to open the room for a group meeting or attend an appointment:

If there's things I need to do like there was giving blood a while ago, I had to schedule a time to go, then I deliberately got it sort of 8:30 in the morning to get myself out of bed. So I had to go there. And then sort of just getting myself out, forcing me to have breakfast and get there and I then find the rest of the day so much better than if I sort of don't have something to force me up relatively early. – William.

Deadlines had a similar effect of helping some people to act, although the combination with stress meant that this had a cost:

I've been in a lot of situations where I pretty much don't have a choice. Like I'll either complete this by the deadline or I'll be homeless sort of thing. That pushes me through, but also makes me live in a constant state of fear. – Margaret.

For others, like Brian, the stress from the obligation outweighed the prompting effect, so that *"a deadline really doesn't help. If anything it makes it harder to start."*

External expectations of another person could not be easily substituted by artificially created structure or urgency using electronic or cognitive strategies. Lists, reminders and alarms were helpful for some; however, when asked if alarms were helpful, Harriet's answer, *"Not really – reminds me I have to do something, but doesn't help me overcome the inertia,"* was typical, as was finding the suggestion laughable. Joel reported that there was *"a big difference between having a support worker and having no support worker,"* and Sam felt the only thing that could help them do things would be another person:

I've pretty much tried everything. We've tried all of that. And it just doesn't, and that's great if it does help for some people but it made me feel there's no hope really. I think unless I have a physical person helping me do these things. – Sam.

Effects on How the Person Feels

General wellbeing, such as energy levels and mental health, both affected and was affected by the participants' initiation impairments. Participants frequently reported being constantly exhausted, stress and overload, difficulty meeting needs, and the effect on (and of) emotions.

Constantly Exhausted

Participants frequently reported states of fatigue which made it harder to act:

My validation has come from my diagnosis which recognized how my extremes of anxiety, uncertainty, executive functioning and SPD [sensory processing disorder] mean I am constantly exhausted. – Jordan.

William found painful emotions so draining he could be unable to act:

I find that sometimes thinking about it then makes me tired because it's [. . .] very painful. But actually, just thinking about it makes me really tired.

Several participants also needed extended periods to recuperate:

It's perfectly valid to me that I'm not doing stuff because I know I'm too tired to do it. I know that it would be stupid to leave the house at this point. I know that it would be ridiculous, I have no spoons[3], as they say, so why am I thinking that I have to do things? [. . .] So I try to be decent to myself. . ..

I know that what I really need is what I call kind of 3-day recoup . . . in which I basically crash for 3 days, watch television, do nothing except eat and watch television and zone out and doze off and stuff. – Emma.

Stress and Overload

Participants talked about stress, both from their lives in general and from the tasks they struggled with. Jackie talked about *"getting so overwhelmed that I can't speak, can't move. My head is just working overtime but I can't actually get any words out."* Often stress was related to sensory aspects of the task or environment, even simply the requirement to move their body.

When people talk about sensory overload, most people assume that just means what they consider to be the main basic senses. I don't think they take into account that stuff such as being too hot or being in too much pain or just being too tired. I don't think a lot of people appreciate that, that just those things can be so overwhelming that it's that difficult to do anything else. – Suzanne.

Stress reduction strategies made things better. Some talked about the benefits of being outdoors or listening to or playing music in order to put them in a better place to approach a problematic task. Stress featured in all of the factors that made initiating more difficult.

Difficulty Meeting Needs

While poor wellbeing made it more difficult to initiate, failing to do things in turn negatively affected wellbeing, creating a self-perpetuating cycle. As described earlier, even basic needs such as drinking or going to the toilet could be left unmet until they became desperate. In addition, many participants described difficulties with exercising and carrying out self-care routines, which affected physical and mental health:

It can also affect my ability of looking after myself sometimes. Showering often enough. Doing my teeth was a massive thing, rarely did them cause I hate the sensory and just process of doing it. Having actual reasons to do things makes a big difference. Just 'looking after yourself' doesn't tend to be a good enough reason. – Helen.

Living conditions also suffered, including difficulty cleaning, clutter, and problems with household tasks. One described her house as a 'constant mess.' Another described various difficulties with maintenance of essential household facilities.

I'll be meaning to buy a tumble dryer for the 5 years I've lived in a house. I've still got paint samples on the wall from when I moved in. My shower was unusable for a year, and I'm lucky to have a bath as well, which doesn't work properly. So, bath's been with a bucket and stuff. – Daniel.

Effects of (and on) Emotions

Strong negative feelings prevented participants getting started on tasks. These included anxiety, painful emotional content, and depression.

[3]'Spoons' here refers to 'spoon theory', a well-known analogy for fatigue and limited resources (Miserandino, 2003).

Lots of the things which I have difficulty would seem to be anxiety driven and based around perfectionism as well. [. . .] For me, it's easier if it's something which I actually feel more comfortable with anyway which is why I think it is anxiety driven. – Suzanne.

However, several participants were clear that although they had anxiety, their inability to do things occurred whether they felt anxious or not:

Even if I feel totally relaxed and happy, you know, some days, I can't formulate the plan so I don't go out at all and that happens once or twice a week. So that is very disabling. – John.

Depressive thinking made Alisha feel she didn't deserve to take care of herself; however, in the light of her recent understanding of autism, she said, *"I wonder these days [. . .] was that depression or was that just this unknown thing that kind of. . . I've never been able to. . ."*

Avoidance of anticipated negative emotions was as important as being paralyzed by overwhelming emotion in the moment.

But the other thing is just that that fear of making it even worse. Because every time I try, it seems to be it seems to end in failure rather than success. And it's just that constant feeling of I messed up again. – Elizabeth.

Emotional factors were mentioned more often as a consequence of failing to act than as a cause:

Anxiety makes it worse, for sure, but I often also have anxiety about not getting things done that I really need to do. – Naomi.

Many participants were able to accept, most of the time, that their inability to act was outside their control, but nearly all expressed frustration as a result. Some felt considerable guilt and inadequacy for their failures, which damaged confidence and made them feel hopeless. This, in turn, made it more difficult to act. Having an understanding that this problem was not something they could control helped considerably.

Sometimes when unable to act, a participant would just get on with something else, while others would be apathetic. The fact that things were not getting done and time was getting shorter would itself often cause frustration, even when the inability to move did not, as discussed in "Disconnection Between Intentions and Actions."

Participants expressed that other people often did not understand the magnitude of their issues. Others would assume that the failure to do a task was due to forgetting and would offer trivial solutions such as alarms and reminders.

People are like, 'well if you just do this, if you set a reminder if you do whatever,' but it's like you have no idea. Like you're so far away from the truth of my existence. I feel like, you make me feel like I'm lying and I end up starting to question my own truth. And I know it's true. – Ruth.

Some were aware that others might see them as lazy or not trying, but recognized that they could not do better.

I know that even if externally to people watching me, it might look like I'm not trying, but I feel like if I'm not getting the stuff done, I know like for myself that it's not just because I'm lazy or not trying. It's just because I can't cope with it at that point, and I can't do it. – Lisa.

Effects on How the Person Lives

When asked about the impact of initiation impairments on their lives, several participants answered that it affected 'everything.' This included both things that they wanted to do and things that they needed to do. Some referred to inertia having a general deleterious effect on their quality of life.

I think I kind of sum it up on my life is probably a lot smaller and less than I would like it to be. Just in general, there's a lot less in it. I would like a bit more in it, but I don't have the ability to make more in it. – Sam.

In addition to the effects on general wellbeing discussed above, there were two further sub-themes describing effects of initiation impairments on their activities: reduced productivity and difficulty maintaining relationships, which were both touched on by Catherine, who said, *"I cannot work and have friends and maintain the house all at the same time. I just can't do it."*

Reduced Productivity

Participants reported that their difficulty doing things affected all areas of productive life; in some cases, the inability to act was the main barrier to employment:

Just my ability to earn money and not relying on the state. And it's just the frustration of, and people meeting you and being like, you're really eloquent and whatever. And it's like so what? It doesn't translate to an ability to utilize that in the world in a way that makes me enough money to live independently and do the things I want to do. – Ruth.

Some found that work was the only thing they could do reliably, although this was always precarious because of the effects on other areas of life.

I'm great at working, but I get stuck on other things. And those things, other things that I'm stuck on eventually become things that affect me, like letting my health decline a little bit. Eventually means I suffer more from stress at work. – Joel.

This difficulty did not only affect things that were aversive or difficult; it also included *"things I want to do and enjoy doing"* (Harriet). They mentioned struggling to start enjoyable work as well as leisure activities such as reading, gardening, exercise, and art:

I'm keen on gardening as some people know but I think since the garden is an allotment it's some distance away that involves me making all sorts of preparations to go out of the door and get there and so I do have some problems in initiating, getting, well, I have problems initiating deciding when I want to go but then I have problems with initiating and getting everything sorted out before I go. – Thomas.

Difficulty Maintaining Relationships

Impact on ability to initiate interaction and maintain relationships was substantial. For some, this was the main problem caused by their inertia. One participant related their

problems with initiating communication, even when it was not anxiety-provoking.

> *I find keeping in contact with people really difficult. I know I should message a lot of people see if they are ok but can't seem to initiate that first message. – Brian.*

Erin was one of several participants who related that, *"All relationships, all friendships in my life only work if the other person is prepared to do a massively disproportionate amount of the initiating, almost all of it."* Some relied on routines, as described in Section "Lack of Voluntary Control," to maintain relationships:

> *I can really only do a friendship where the other person is willing to commit to seeing me on a regular schedule, like we'll always see this day or whatever [. . .] And even if it's just well they've gone, 'well let's just agree that we will try and see each other once a month.' Well they can't do that. I can do once a week and I can do once a week and maybe some weeks we try but we can't do it, but I can't do [once a month]. – Alex.*

Relationships could also be strained by others' difficulty understanding why the autistic person was not getting more done. The judgment of others could make the difference between acceptance and a negative experience:

> *I am not sure it's ALWAYS negative. . . Like sometimes I just embrace it and go with it and accept that that's a month where I'll be in one corner of the sofa, eating junk food and playing video games, because that's all I can manage. It becomes negative when I try to force myself out of it or put it under the scrutiny of external judgment from other people. – Margaret.*

DISCUSSION

This study is a broad preliminary investigation into the experience and impact of autistic inertia. It arises from the concerns of autistic people, including the lead author, some of whom have said that this is the most disabling aspect of their autism (Murray, 2017). This study is unique in considering difficulty with tasks of any type, not exclusively social, and by specifically looking at difficulty acting on intentions, rather than sensory or motor experiences more broadly. Furthermore, this study gathers focused qualitative data directly from autistic people who are able to describe their own experiences. From those descriptions, we have found that difficulty acting on intentions arises from associated tendencies to resist stopping, starting and changing activity. While difficulty with planning and prioritizing was common, a subset of participants described a more profound impairment in initiating even simple actions. Participants described complex interactions between various external and internal factors and their ability to act. What was consistent and universal among our participants was that the inability to start and stop activities at will had profound and pervasive effects on their day-to-day lives and general wellbeing.

Characteristics of Initiation Impairments

The first goal of this research was to document the difficulties that autistic people experience acting on their intentions, which will both help in understanding these impairments and point out possible avenues for further research. The characteristics of these initiation impairments will be considered in terms of emotion and motivation, executive function, and movement.

Emotion and Motivation

While autism is now recognized as a neurological condition, there is still a tendency to view autistic behavior as social, emotional and volitional rather than the manifestation of a differently functioning brain. Too often, autistic people are considered non-compliant or unmotivated when they fail to act. It would be easy to attribute their inaction to laziness or lack of motivation; however, several characteristics of autistic inertia distinguish it from voluntary task avoidance. First, while one may procrastinate about doing a chore that is aversive, inertia also affects activities the person enjoys. Second, even for tasks that are difficult or unpleasant, a strong enough motivator can activate an avoidant person. By contrast, participants in our study could not overcome their inertia in order to carry out a task that was important to them, often even those driven by basic needs. Third, our participants experienced as much difficulty stopping as starting, so they were not simply avoiding effort. And finally, rather than enjoying their diversion from an undesirable activity, our participants were often frustrated, annoyed and even physically uncomfortable due to their inability to act. While transient lack of motivation and avoidance of undesirable tasks is a normal part of life, this debilitating level of initiation impairment affecting even simple and enjoyable actions is clearly beyond the typical experience.

There are several possible explanations for these experiences, aside from avoidance or non-compliance. For example, negative emotions and inaction were connected in a self-perpetuating cycle, where failing to do things created bad feelings which, in turn, made it more difficult to act. These factors are not unique to autism, but depression and anxiety occur at high rates in autistic adults (Hollocks et al., 2010; Hudson et al., 2018) including our participants. Nevertheless, our findings highlighted that initiation impairments cannot be entirely explained by motivational or emotional factors. Where anxiety did feature, it was not always clear whether it was causal; sometimes it seemed as if the person assumed anxiety was the cause because they could find no better explanation for failing to act.

For our participants, the most profound episodes of being 'stuck' were also the least likely to be connected with strong emotion. Catatonia-like physical freezing was often accompanied by blunted or absent thoughts and emotions. Although stress and anxiety could make episodes more likely, the overwhelming anxiety or depression reported by others (Paterson, 2016), such as being 'frozen with fear' or deeply unmotivated due to low mood, were not a proximal feature of these episodes, which were more often characterized by emotional detachment. Their lack of emotional arousal was remarkable given that they were often conscious of mounting discomfort (e.g., thirst and pain) and unpleasant consequences of failing to act. During such episodes, our participants also often experienced altered awareness of self, the environment and the passage of time. This was distinct from being absorbed in an activity where they may 'lose track of

time' in that the person had reduced or absent ability to initiate voluntary movements. They often felt disconnected from their body and actions in a way that resembled dissociative experiences (Ben Shalom, 2000). Dissociation is associated with stress and trauma, an area of increasing interest in autism research (Brenner et al., 2018), and more than one third of our sample reported a current or past diagnosis of Post-Traumatic Stress Disorder. Further investigation is indicated to clarify any relationship between dissociation and the detachment experiences described by our participants.

Participants described with poignant clarity the profound impact of these difficulties, which have so far escaped the notice of most autism researchers and clinicians. More fundamentally, the incomprehensibility of *why* they didn't 'just do things,' affected their self-concept as capable people. As autistic blogger, Sparrow (2016), writes, "it's hard not to feel lazy or inadequate about one's own inertia without the proper understanding of what it really is and what it really means." The ability to respond to one's environment at will is intimately connected with social interaction, agency, and identity.

Executive Function

Rather than an emotional basis, it is possible that difficulty initiating can be an outcome of executive dysfunction. Executive function is a diffuse concept with highly varied profiles found in previous research with autistic people (Demetriou et al., 2018). Flexibly starting, stopping and switching tasks depends on executive function (Hoofs et al., 2018; Yeung and Chan, 2020). Some of our participants had difficulty breaking down a task, but more often they broke it into so many components that it became overwhelmingly complex and impossible to find the starting point. This tendency to excessively segment a task may be a manifestation of autistic detail orientation (Mottron et al., 2006). The ease with which participants could be derailed from an activity may suggest a weakness in working memory or high distractibility. Those who experienced problems with sequencing a task benefitted from help finding the first step, which is consistent with experimental research finding an initiation-specific executive function impairment that could be overcome by providing the first step (Carmo et al., 2017). Prior research has investigated executive functioning deficits in autism, but our research is unique in considering this from an autistic perspective in an ecological context, which highlights the profound impact on accomplishing tasks in everyday life.

Difficulty switching between actions can be problematic when stuck in an inactive state, but a strong fixed attentional focus, sometimes referred to as 'monotropism' (Murray et al., 2005), also facilitates highly productive periods and a deep immersion in nature and hobbies. This experience is similar to 'flow states', which autistic people may experience from atypical sources, such as when engaging with specialist interests (Milton, 2017). Our participants occasionally experienced paradoxical bursts of high levels of productivity. These periods were described both as enjoyable immersive flow states and as panic-driven hyper-productivity. Such focused immersion can become problematic when it is so intense that it overrides shifting attention to other necessary or desired tasks. Nonetheless, in itself, a narrow focus is a natural and non-pathological aspect of autism, and attempts to overcome inertia by teaching the autistic person to be more flexible or engage in more varied activities would be misguided. The difficulties of autistic inertia need to be supported so that the related positives can be fully appreciated. Further research is needed to clarify the nature of the relationship between autistic inertia, resistance to change, and intense focus.

Movement

Rather than being primarily a cognitive, emotional or social deficit, both the failure to act and the lack of response to that failure could at times be due to an impairment of voluntary motor initiation. This difficulty shares characteristics with those of autistic catatonia described by Shah (2019), as detailed in **Table 3**, but often more subtle. This type of initiation difficulty is manifest as a loss of conscious voluntary control of goal directed action affecting even simple, familiar actions such as standing up from a seated position or reaching for a drink. A further distinctive characteristic of this type of experience is the response to interruptions. When experiencing inertia characteristic of executive function impairments, interruptions were perceived as an annoyance and avoided if possible as participants found it difficult to return to the original task. Conversely, when in a disconnected catatonic state, a small interruption such as a noise from another person could trigger an end to the episode of immobility.

Due to the limited communication abilities of those affected, the existing literature on catatonia is entirely by carer report and observation. In one such study, Breen and Hare (2017) found difficulty initiating actions was the least common of six 'core' catatonia symptoms. However, as intentions are invisible, and a lack of emotional arousal and an intact ability to make other movements could mask the unrealised intention to move, difficulty with initiation may be underestimated by carer reports. For this reason, such phenomena can only be fully explored through the subjective experience of autistic individuals. Broader experiences of autistic embodiment, including motor and arousal control, have been explored through first-hand accounts by autistic people (Welch et al., 2018, 2020). In these accounts, autistic people also report a variety of difficulties with controlling their action and inaction, including feeling a 'mind-body disconnect.' By specifically asking autistic participants about such episodes, the present study provides unique insight into the internal experience, and the ability of our participants to fully articulate these experiences may be further enhanced by their connections to and interactions with the autistic community.

While there is some value in considering different approaches to initiation problems that have a primarily emotional, executive function, or movement profile, these are not completely dissociable. The association between anxiety and catatonia (Shah and Wing, 2006, p. 250) is inconsistent, with some studies reporting high levels of anxiety in up to 80% of catatonic patients (Northoff, 2002) and others reporting none (Pelzer et al., 2018). Furthermore, impairments in executive function, movement and motivation (variously called apathy, avolition or initiative impairment depending on the area of study) co-occur in a variety of neurological and

psychiatric conditions including parkinsonism, depression and schizophrenia (Yamanaka et al., 1996; Ozonoff and Jensen, 1999; Bertilsson et al., 2018). Given these associations and the overlapping cortico-striatal circuitry involved in cognitive flexibility and movement control (Daniels, 2009; Uddin, 2021), these may be compatible rather than competing explanations. Teasing these apart and specifying the relationship between them is beyond the scope of this paper but should be explored in further research as they may lead to understanding of the mechanisms and interventions for the most debilitating of autistic initiation impairments.

Implications for Understanding and Supporting Autistic People

Autism is currently characterized as a dyad of impairments in (i) social interaction, and (ii) flexibility (American Psychiatric Association, 2013). A prominent finding which may be surprising to those who view autism as primarily defined by social deficits was that difficulty maintaining relationships was one of the most frequently mentioned negative impacts of their initiation impairment. Contrary to the view that autistic people initiate interaction less often because they are less interested in others (Chevallier et al., 2012; Kohls et al., 2012), participants in our study wanted to contact people who were important to them, but found themselves unable to initiate. In respect to the other aspect of the core dyad, our research suggests that resistance to change relates not only to repetitive motor mannerisms and resistance to transitions imposed by others, but also with starting and stopping internally motivated actions.

The experiences described in the 'disconnection between intention and action' theme support the view of a small number of researchers who propose that many autistic characteristics may be attributed to sensorimotor differences (Robledo et al., 2012; Donnellan et al., 2013; Torres and Donnellan, 2015). Understanding the role of various factors underpinning difficulty initiating action can enable more successful support strategies. The core characteristics of inertia and answers to the focus group question 'what helps' have led to some principles to consider when trying to assist an autistic person struggling to initiate tasks, which are described in **Table 4**.

Participants almost universally found that conventional organization and memory tools such as alarms, lists, reminders and calendars were seldom helpful; practical assistance was far more beneficial. Initiation impairments were often related to the height of the cognitive threshold to overcome, so it was more difficult to get out of bed than to pick up a phone, and complicated activities such as leaving the house were especially difficult. Having another person provide all necessary information or start off the task lowered the initiation threshold, thereby facilitating action.

Social connections were not only one of the most significant casualties of their impairments, but also very important in mitigating the effects of initiation impairments. Prompting from another person in their presence was the most helpful intervention. Even having someone working nearby without interacting was often helpful. This is consistent with evidence from executive function research (Williams et al., 2014) and similar effects have been reported in books on catatonia by Sacks (1976) and Shah (2019, p. 108). Participants also found it easier to do anything where another person was depending or counting on them, even from a distance, and most difficult to do something only for themselves. Mistimed or misdirected prompts or excessive demands and pressure could cause stress which would exacerbate issues. This also applied to internally generated pressure such as self-imposed 'deadlines' and schedules. Several participants had developed personal techniques to reduce the pressure of expectation. For example, by telling themselves *"all you have to do is..."* one tiny step, they could circumvent the sense of pressure and demands that could cause them to get stuck.

TABLE 4 | Principles for helping with autistic inertia.

Principle	Explanation and examples
Distinguish between mechanisms Consider whether the current difficulty acting is underpinned by motivational/emotional, organizational or movement problems, because they have different responses to support.	• *Motivational:* tasks that are stressful, aversive, or anxiety inducing. • *Organizational:* tasks that are complex or involve transitions. • *Movement:* can affect even very simple tasks and meeting basic needs.
Use continuity When the autistic person wants to continue with a task, make it easy to continue.	• Avoid interruptions, e.g., provide all information necessary to make a decision at the time the question is asked. • Avoid unnecessary transitions and interruptions. • Keep moving, e.g., avoid sitting down between active tasks.
Use prompts carefully Prompting can be helpful, but if used incorrectly can exacerbate difficulties.	• Sensitively delivered without adding stress. • During natural breaks in attention. • To break away from disconnected passive states. • Avoid nagging to attend to others' priorities as such demands are stressful and exacerbate issues.
Environmental scaffolding Provide an environment that supports action	• Do tasks in an environment specific to those activities, e.g., working in a designated study or office. • Engage in compatible activity nearby. • Keep a regular routine.
Lower the threshold Make it easier to start by lowering the initial hurdle	• Self-talk or encouragement to only do one small step in the desired direction. • Have someone else do the first step.

Outline of five key principles to apply the results of this study to assisting autistic people to initiate tasks. Five key principles (bold) to apply the results of this study to assisting autistic people to initiate tasks. Each is accompanied by a brief description and examples of practical applications where appropriate.

Limitations and Next Steps

While providing novel insights, this study is limited by the inherent limitations of a broad research question. This has not allowed for detailed inquiry about the influence of gender, co-occurring diagnoses, personal history, or other differences. We hope that this research will inspire others to look at autistic inertia further, including these nuances.

The participants in this study should not be considered representative of the autistic population as a whole. The focus

on internal experiences excludes those who are unable to reflect on or express their experiences in an accessible way. Although Welch et al. (2018) found overlapping themes in the memoirs of minimally verbal autistic young people, the experiences of those who do not use the written or spoken word to communicate remain inaccessible to this form of enquiry. Those who attended the face-to-face groups are more likely to be sociable and to tolerate participation in groups. This was offset by also conducting text-based online focus groups which included people who were unable to attend Autscape or participate in a verbal group interaction. Our participants may also have been more likely to attend if they experienced difficulty with inertia, as the purpose of this study was to describe the phenomenon rather than to draw inferences about prevalence.

Recruitment of a majority of participants from the Autscape event also limits the range of the sample. Autscape participants are more likely to be introspective about their experiences of autism and to have communicated about them with other autistic people. While this limits the representativeness, it is also an advantage for this early enquiry. One of the goals of this study is to provide language to express experiences of being unable to act. By drawing from a community where autistic people share and develop their understanding of autism, they are more likely to have developed ways of reflecting on and expressing their experiences and strategies to overcome difficulties. The text-based focus groups included participants who had never attended Autscape, yet the themes were very similar, with no novel themes arising in these groups.

Possible future directions include exploring experiences of autistic inertia in the context of gender, living circumstances, support needs and co-occurring diagnoses, which were not considered in the current study. Although there has been some quantitative research characterizing executive deficits in autistic people, further research is needed to understand the impact of these on day-to-day life, including aspects of inertia such as those described by the 'difficulty finding the first step' and 'tendency to maintain one state' themes and possible overlap with ADHD traits. Furthermore, the present study has indicated directions for investigation into possible associations with stress, dissociation, avolition, catatonia, and the possible underlying cortico-striatal circuitry. An increased understanding of these may help to tease apart the different mechanisms, improve understanding of these issues, and begin to work toward helpful interventions.

The lead researcher's personal experience of severe initiation impairments suggestive of a movement disorder, and her personal interest and prior informal investigation of the topic may have colored interpretation of the data. However, this author's personal interest and experience has also been an asset in understanding the issues and building rapport with participants. Future research should continue to adopt a participatory research framework.

This research was prompted by members of the autistic community who experience the disabling aspects of inertia. The absence of documented evidence that difficulty initiating action is part of the autistic experience hampers access to understanding and effective support. While the focus of interventions for autistic people is on anxiety and social issues, many supporters are not even aware that inertia can be 'the single biggest problem' (Murray, 2017) arising from autism, creating a life that is *"a lot smaller and less than"* (Sam, focus group) it should be.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by University of Manchester Research Ethics Committee. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

KB conceived the idea and designed the investigation with support from EG, KL, and EP, who supervised the project. KB collected and analyzed the data and drafted the manuscript. All authors discussed the results and provided critical feedback which helped shape the research and analysis. All authors contributed to the final manuscript.

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A Subtle Profile With a Significant Impact: Language and Communication Difficulties for Autistic Females Without Intellectual Disability

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The presentation of autism in females is poorly understood, which is thought to contribute to missed or later- age diagnosis, especially for those without intellectual disability. Dedicated research into social and behavioral differences has indicated a specific female phenotype of autism. However, less has been done to explore language and communication profiles, despite known sex/gender differences in typically developing populations. This article provides a synthesis of recent work from this small but emerging field. It focuses on a series of four preliminary and explorative studies conducted by the authors and embeds this within the wider literature. Findings suggest a specific profile of language and communication strengths and weaknesses for autistic females without intellectual disability (compared to autistic males and typically developing females). Furthermore, despite the relatively subtle presentation of difficulties (compared to autistic males), the impact on functionality, social inter-relations and emotional well-being, appears to be equitable and significant. The discussion highlights the need for further empirical research and proposes areas for investigation. Implications for clinical practice include the need for better recognition, testing and provision of interventions dedicated to the language and communication difficulties for autistic females. This has relevance for diagnostic, mental health and speech and language therapy services.

Keywords: autism, language and communication, sex/gender differences, social impact, emotional impact, functional impact

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INTRODUCTION

Sex/gender[1] differences in language and communication profiles for typically developing individuals are well documented in the literature. Females demonstrate earlier acquisition of first words (Bleses et al., 2008), better and earlier integration of language with gesture (Eriksson et al., 2012), earlier examples of social-emotional vocabulary (e.g., "like," "please"), and use of more complex linguistic forms during spontaneous speech (Bouchard et al., 2009). They also use language and communication differently from males, focusing on person-centered topics and emotions (Newman et al., 2008), and using collaborative and negotiated discourse

[1]The term "sex/gender" is used to reflect the understanding that individuals' identities are composed of hard to distinguish features of biological "sex" and socially constructed "gender."

(Ladegaard and Bleses, 2003). Importantly, this profile appears to be expected within interactions (Newman et al., 2008) and is linked to successful integration with female social groups (Tierney et al., 2016).

Sex/gender differences in autism have received growing attention in recent years, although this has focused on social and behavioral domains rather than language and communication. Currently females are diagnosed in lower numbers (1:3) than males (Loomes et al., 2017) especially in groups with higher cognitive function (1:7; Nicholas et al., 2008). This is despite autistic symptomatology existing with relative parity (2:1) in whole population samples (Giarelli et al., 2010). Clinical concerns are that females are being missed from diagnosis due to poor recognition of the autistic female phenotype (Kreiser and White, 2014). Sex/gender differences have been identified in rigid/repetitive behaviors using diagnostic measures (Van Wijngaarden-Cremer et al., 2014; Hull et al., 2017a) with males typically exhibiting increased frequency and severity compared to females. Differences in social interactions have been better identified using specific measures, avoiding the homogenizing effect of collecting data and constraining participant groups using the same diagnostic tools (Lai et al., 2015). Several studies now point toward a distinct profile of social-interaction difficulties for females compared to males, using measures of empathizing (Rieffe et al., 2021), friendship (Sedgewick et al., 2016), play-behaviors (Dean et al., 2014), and emotional reciprocity (Head et al., 2014). A review of the literature found little evidence of language and communication differences between sex/gender in autism (Hull et al., 2017a). However, data in those studies were collected using isolated measures (parental reports or basic vocabulary tasks), where difference may be under-identified for reasons discussed in this paper. Others used diagnostic measures, which may incur a homogenizing effect by constraining participants and measuring difference using the same tools (Lai et al., 2015). This current article focuses on the smaller body of work investigating subtle sex/gender difference using specific measures of language and communication, in pragmatic and above sentence-level language. Principally, it will consider four clinically driven studies from the authors' research group; using direct assessment (Sturrock et al., 2019b), observation and report measures (Sturrock et al., 2019a), child interviews (Sturrock et al., 2021) and parental interviews (Sturrock et al.,), and synthesizes these with recent findings from the wider literature. It proposes that autistic females most likely to be missed from diagnosis (those without intellectual disability: IQ $\geq$ 70) have a specific profile of language and communication skills, different from both autistic males and typically developing females, and that these differences make them prone to negative social, functional and emotional sequelae. It calls for further research and proposes areas for investigation.

ASSESSMENT OF A SUBTLE PROFILE OF DIFFICULTIES

While subtle language and communication differences are identified between autistic individuals (without intellectual disability) and typically developing (TD) controls (Howlin, 2003; Kelley et al., 2006), this is rarely achieved through basic structural language assessment (e.g., testing vocabulary and sentence-level grammar). Neither is basic structural language expected to differ between school-aged and above TD females and males (Newman et al., 2008). An attempt to explore sex/gender difference must therefore utilize measures with the capacity to compare subtly differing profiles.

Sturrock et al. (2019b) proposed a battery of direct assessments targeting language (expressive and receptive) at multiple levels (word, sentence and above sentence-level/narrative), word knowledge (semantics), inference and vocabulary of emotion. In subsequent work, the authors proposed a series of functional communication measures (Sturrock et al., 2019a) including parent and child questionnaires and observational checklists for social use of language (pragmatic skills). Details of assessment measures are found in **Supplementary Appendix 1**. These measures were undertaken with a cohort of 52 children without intellectual disability in a 2 (diagnosis: Autism/TD) by 2 (sex/gender: female/male) design. Children were recruited from a narrow age range (8y11m–11y6m), to minimize the effect of increasing language abilities across development. Children in middle childhood were purposefully selected, being young enough to avoid interference of secondary mental health conditions (social communication difficulties are thought to increase in secondary school for autistic girls; 6) but old enough to be post- diagnosis (likely to occur much later for autistic girls (Rutherford et al., 2016). Overall, participants had PIQ $\geq$ 70, and there were no statistical differences on basic vocabulary and grammar skills or autism severity between groups (see **Supplementary Appendix 2**). **Figure 1** provides a depiction of assessment measures per child.

As predicted from the literature (Howlin, 2003; Kelley et al., 2006) no group differences were identified in receptive or expressive vocabulary or sentence-level language. However, it is possible that other measures may have provided a more discrete assessment of difference; for example, The Index of Productive Syntax (Scarborough, 1990) showed group differences in expressive sentence-level grammar when comparing spontaneous language samples of autistic children without learning disability and TDs (Eigsti et al., 2007). Similarly, subtests for following oral instruction within the CELF (Semel et al., 1987) and NEPSY (Korkman et al., 1997) assessment batteries, demonstrated problems in receptive ability (Koning and Magill-Evans, 2001; Saalasti et al., 2008) for autistic children without intellectual disability compared to controls. Sex/gender differences in these language subtests have not been explored but may have better capacity for identifying subtle variations and are worthy of investigation. Another consideration is the existence of heterogeneity amongst autistic individuals and the probable existence of a subgroup with specific grammatical language impairment (Roberts et al., 2004; Wittke et al., 2017). Similar to the non-autistic population specific language difficulties can occur in autism without other intellectual disability, the prevalence of this within autistic girls is currently unknown. In larger population studies, it would be important to isolate this group for separate consideration in analysis. The findings

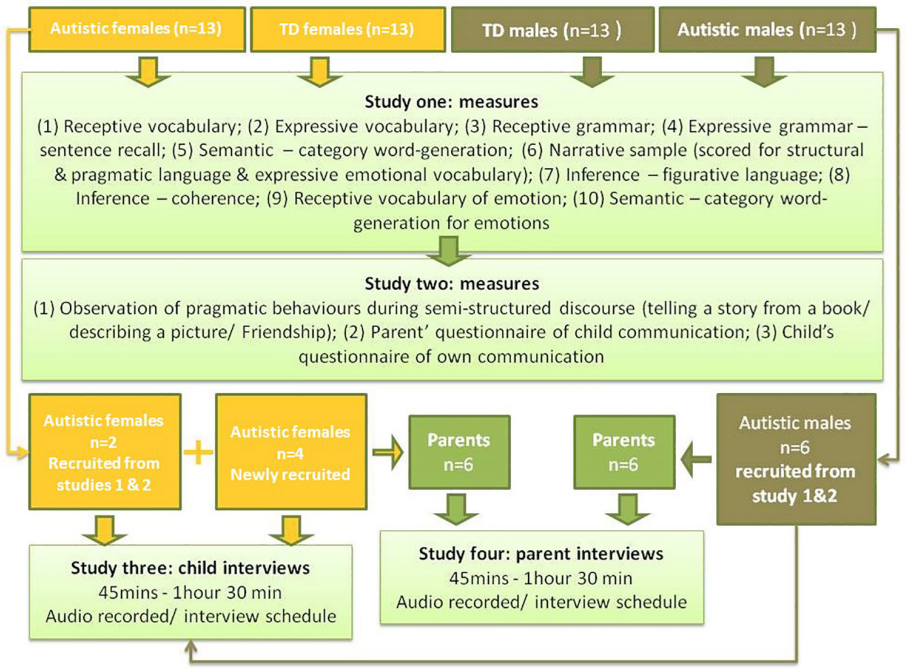

FIGURE 1 | Flow diagram of participants and measures over four studies of language and communication.

from the author's series of studies focuses on the profile of autistic girls without such additional and specific grammatical difficulties, as evidenced by the children's performance on the basic structural language tasks.

SEX/GENDER DIFFERENCE IN NARRATIVES

Narrative has been used to demonstrate subtle deficits in the language and communication skills of autistic individuals without intellectual disability, even when basic structural language is in normal range. Narrative requires the individual to recall, organize and present information in a way that orients the listener to story meaning; blending cognitive and linguistic skills (Norbury et al., 2014) with an ability to interpret social cues from the listener (Volden et al., 2017). Mixed-sex/gender or male autistic groups without intellectual disability have demonstrated deficits in structural (Diehl et al., 2006; Rumpf et al., 2012; McCabe et al., 2013) and pragmatic (Capps et al., 2000; Losh and Gordon, 2014; Banney et al., 2015; Kauschke et al., 2016) features of narrative. It therefore provides scope for demonstrating differences in higher-level language and communication profiles and potentially between sex/gender.

Sturrock et al. (2019b) found autistic females and males performed similarly but behind TDs in their use of temporal connectors ("and then.") and number and range of causal connectors ("so.") leading to overall limitations with structural complexity and pragmatic coherence. This may potentially support the argument for subtle group differences in higher-level linguistic competency (Kelley et al., 2006; Eigsti et al., 2007; Saalasti et al., 2008). Other studies have demonstrated sex/gender differences in pragmatic elements of narrative, with autistic females generating richer character depictions and descriptions of internal states, cognition, perception and judgment (Kauschke et al., 2016; Boorse et al., 2019; Conlon et al., 2019) and overall better skills in retelling salient story elements (Conlon et al., 2019). When compared to typically developing peers, however, autistic girls experienced difficulties on these measures (Kauschke et al., 2016). Sturrock et al. (2019b) also found autistic females and males performed behind typically developing children in their use of vocabulary of emotion in narrative. These relative difficulties for autistic girls compared to TDs may put them at a functional disadvantage in terms of social integration (Dean et al., 2014) and self-advocacy (Sillar et al., 2014). The need for integrating linguistic information with social cues (Volden et al., 2017) may explain better outcomes for autistic females on pragmatic elements of narrative. This may be grounded in other noted advantages for females; in social motivation (Head et al., 2014; Sedgewick et al., 2016) and social attention (Harrop et al., 2018). It would therefore be of interest to isolate underpinning linguistic and socio-cognitive skills in narrative and investigate the influence of sex/gender on those.

SEMANTIC SEX/GENDER DIFFERENCES

Sex/gender difference in this language and communication domain are particularly poorly investigated, despite being one

of the more widely recognized linguistic impairments in autism more generally (Groen et al., 2008). However, Sturrock et al. (2019b) and Goddard et al. (2014) found that autistic females performed better than autistic males using similar word-generation/fluency tasks. They also both found that autistic girls performed behind TDs on the same measures. Sturrock et al. (2019b) asked participants to name as many words as they could from four categories (animals, food, occupations and emotions) within a 60-s limit. Raw scores for "animals," "food," and "occupations" were amalgamated into one composite score and analyzed separately from the category "emotions." Unlike expressive vocabulary tasks (like the TOWK), word-generation tasks require the individual to generate multiple word examples from a single category (relying on a flexible interpretation of word meaning) and does not provide visual stimulation to aid recall. These features may explain why semantic/word-generation tasks are more commonly occur in autism (Groen et al., 2008) while expressive vocabulary may be unimpaired. Secondary analysis in Sturrock et al. (2019b), study suggested that the sex/gender differences occurred within categories as well as using the composite score. Autistic boys demonstrated relatively elevated performance in the category of "animals" which observationally was associated with specialist knowledge in this area (typified by low-frequency, highly specialist exemplars; lion-mane jellyfish, stork-eyed beetle, goblin shark). The interaction between special interests and vocabulary acquisition is an area of potential future research, which might help explain elevated idiosyncratic word choices reported in autistic groups (Walenski et al., 2008). Further, differences in performance on semantic category word-generation tasks have been associated with differences in lexical organization between autistic and non-autistic groups (Gaffrey et al., 2007), highlighting the need for investigations of sex/gender differences in mechanisms of the development of semantic organization and their relationship to outcomes on these tasks.

SEX/GENDER DIFFERENCES IN PRAGMATICS: INFERENCE AND DISCOURSE BEHAVIORS

Inference is identified as a persistent difficulty for autistic individuals without intellectual disability (Loukusa and Moilanen, 2009), relying on core language (Tzuriel and Groman, 2017) and social-cognition skills (Martin and McDonald, 2004). Currently, there is very limited investigation into sex/gender differences in pragmatic inference. Two tasks in Sturrock et al. (2019b) provide some early insight: one interpreting meaning from figurative language (MacKay and Shaw, 2004), the other interpreting coherence within text using world knowledge (Jolliffe and Baron-Cohen, 1999). The children were asked to explain speaker's intended meaning and demonstrate meta-awareness of a range of figurative language examples in the first task, then asked to identify missing information implied within a short story in the second. These early investigations suggested that autistic females perform better than autistic males and worse than typically developing females on tasks requiring inferential interpretation. Further investigation is of course required. However, it is in keeping with the literature that underlying skills in social awareness may put autistic females at an advantage on these tasks. These early findings suggest important differences in inference between autistic females and males, with consequent implications for diagnosis. They point to fruitful further work investigating sex/gender difference in other measures of inference, and highlight the importance of isolating the relative impact of social cognition or linguistic ability on performance.

By contrast, sex/gender differences in pragmatic behaviors during discourse have had more attention in the wider literature. Sturrock et al. (2019a) used the Pragmatic Rating Scale (PRS; Landa et al., 1992) as a measure of observable pragmatic features within semi-structured discourse (using the Autism Diagnostic Observation Schedule-Second Edition; Lord et al., 2012). Total PRS scores (Sturrock et al., 2019a) again showed autistic females performing better than autistic males but behind typically-developing females, replicating the pattern found in pragmatic (inference) tasks (Sturrock et al., 2019b). Differences were driven by performance on discourse management, communicative use of speech and language and non-verbal skills. Although specific analysis of sex/gender differences in discourse have not yet been undertaken, they will certainly have an important impact on the social experiences of autistic individuals. For example, Cola et al. (2020) found autistic females performed better than autistic males on a measure of first impressions during naturalistic conversations. The authors proposed first impressions would be based on judgments of pragmatic behaviors such as vocal prosody, gesture, facial expressivity and general awkwardness, although this was not expressly tested. Similar findings occurred during observation of video-recorded interactions in a study by Cage and Burton (2019). Better conversational reciprocity for autistic females compared to autistic males was also identified using diagnostic criteria in DSM-IV and DSM-5 (Hiller et al., 2014) and through analysis of appropriate pause markers, e.g., "um" as opposed to "uh" during speech samples (Parish-Morris et al., 2017). It has been suggested that this could be associated with females' masking of autistic features (Parish-Morris et al., 2017), a phenomenon associated with camouflaging autistic behaviors more generally (Hull et al., 2017b). However, pragmatic language requires skills which integrate linguistic content with social context (Baird and Norbury, 2016), and as previously described autistic females' elevated outcomes on social measures (compared to autistic males) may be due to natural differences in social attention and motivation (Head et al., 2014; Sedgewick et al., 2016; Harrop et al., 2018). Detailed discourse analysis could contribute to better understanding of subtle differences in conversational behaviors between autistic females and males and should be compared to normative data.

SUBTLE PROFILE AND SIGNIFICANT IMPACT

Overall, then, early findings suggest that autistic females will present with a subtle profile of language and communication difficulties compared to autistic males, yet they continue

to demonstrate difficulties compared to typically developing females. This mirrors findings from research into social interactions (Sedgewick et al., 2016) and play behaviors (Knickmeyer et al., 2008). Their subtle presentation, compared to autistic males, may easily confound diagnosis, limiting access to appropriate services and indirectly leading to poorer functional outcomes and emotional well-being (Bargiela et al., 2016). However, it is also important to consider whether fewer language and communication difficulties as measured by direct assessment, will equate with fewer *perceived* difficulties as reported by the individual or their parent.

The limited data appear to suggest that when asked to rate language and communication difficulties autistic females and their parents perceive a similar level of deficit as autistic males and their parents (Sturrock et al., 2019a). This was shown using the CC-SR (Bishop et al., 2009), and CCC-2 (Bishop, 2003). This may indicate equal levels of perceived difficulties experienced by autistic females and males.

Although hard to interpret, similar findings were identified when autistic individuals (Holtmann et al., 2007) and their parents (Lai et al., 2011) were asked to rate their autism severity. As with the language and communication data, females and males perceived their levels of difficulty to be equally severe, despite females presenting with lower severity on more objective measures of clinical observation. It has been hypothesized that this phenomenon is related to the higher social expectations placed on females (Holtmann et al., 2007), meaning their reduced level of difficulty could be offset by an increased level of demand. It could also demonstrate that autistic females and their parents are acutely aware of subtle functional difficulties when compared to typically developing peers, a disparity reflected in the comparative data already discussed (Knickmeyer et al., 2008; Sedgewick et al., 2016; Sturrock et al., 2019b).

Therefore, despite a relatively subtle presentation of language and communication difficulties, autistic girls and boys without intellectual disability might be expected to experience a similar level of impact. Detail of that impact was provided in qualitative accounts (Sturrock et al., 2021) from 12 autistic children (6 girls, 6 boys). Daily living (participation and self-advocacy), social interrelations (social interactions and relationship-building) and emotional wellbeing (reactive and longer-term negative emotions and difficulties help-seeking) were all identified as areas of direct impact. Preliminary analysis of parental interviews ($n = 12$) seems to support these assumptions (Sturrock et al., in preparation). **Supplementary Appendix 3** provides details of interviewee characteristics.

Thematic analysis found that difficulties with discourse, listening and word-finding were strongly associated with breakdown of conversations. These may contribute to results from recent empirical research, which suggests language difficulties will predict poorer social performance in autistic individuals (Levinson et al., 2020). Additionally, the associated effort incurred in managing these difficulties often resulted in avoidance or limitations to social participation. In child accounts, narrative difficulties were closely associated with limitations in explaining events, thoughts and ideas, and this in turn was related to difficulties with self-advocacy and social integration, as predicted in the literature (Dean et al., 2014; Sillar et al., 2014). **Supplementary Appendix 4** shows a representative sample of quotes and themes.

These subtle difficulties experienced by autistic girls were also commonly associated with feelings of frustration, anxiety and negative sense of self-worth. The negative impact of communication difficulties on mental health are recognized in non-autistic populations (Levickis et al., 2018), but less well explored in the autism literature. This is an area of particular interest for future research due to the higher rates of associated mental health conditions in autistic individuals without intellectual disability (Leyfer et al., 2006).

The children interviewed not only described a negative emotional impact from communication difficulties, they (and their parents) also reported specific difficulties expressing emotional content in personal narratives. Recognition of emotion is thought to be limited in autistic individuals (Uljarevic and Hamilton, 2013) and this may be linked to underpinning difficulties with social cognition for the group (Löytömäki et al., 2020). However, recent research suggests that relative to autistic males, autistic females may be more inclined to comment on the emotions of others (Rieffe et al., 2021), they may have better skills in recalling emotional memory (Goddard et al., 2014), more advanced receptive and expressive use of vocabulary of emotion (Sturrock et al., 2019b) and improved narration of the internal states of others (Conlon et al., 2019; Kauschke et al., 2016). As emotional literacy is linked to better well-being (Eisenberg et al., 2005) through support-seeking and self-regulatory mechanisms, its relationship with sex/gender and communication difficulties is an important area of research interest.

DISCUSSION AND FUTURE DIRECTIONS

This overview of the current literature strongly suggests that language and communication difficulties present differently for autistic females without intellectual disability, compared to autistic males with the same IQ and autism severity. This may contribute to poorer recognition and lower diagnostic rates of autism in this group. Areas of greatest sex/gender difference appear to exist in domains where meaning of structural language is mediated by social context; inference; language of emotion and internal state; and pragmatic behaviors (discourse and pragmatic features of narrative). See **Table 1** for an overview of those findings.

Female advantages in pragmatic and semantic tasks may be linked to natural advantages in social motivation and attention, when compared to autistic males. This interaction should be explored and compared to the influence of higher-level linguistic skills.

Fewer studies provide sex/gender norms but where they do exist, autistic females appear to perform behind typically developing females on measures of pragmatics, semantics, and

TABLE 1 | An overview of key findings showing a comparison between autistic females, autistic males, and TD females.

Measure and paper	Autistic female compared to autistic male	Autistic female compared to TD female
Basic structural language direct assessment study one: Sturrock et al., 2019b		
(1) Receptive vocabulary (2) Expressive vocabulary (3) Receptive grammar (4) Expressive grammar		
Semantic direct assessment study one: Sturrock et al., 2019b		
(5) Semantic category (word generation)		
Narrative direct assessment study one: Sturrock et al., 2019b		
(6) Narrative (structural language)		
(6) Narrative (pragmatics/coherence)		
Inference direct assessment study one: Sturrock et al., 2019b		
(7) inference (figurative language)		
(8) inference (coherence)		
Language of emotion direct assessments study one: Sturrock et al., 2019b		
(9) Receptive vocabulary of Emotion		
(10) semantic category (word generation: emotion)		
(6) Narrative (expressive vocabulary of emotion)		
Functional language and communication measures study two: Sturrock et al., 2019a		
(1) Observation of pragmatic behaviours during semi-structured discourse		
(2) Parent's questionnaire of child communication		
(3) Child's questionnaire on own communication		
		

Based on mean averages from a range of measures across two studies Sturrock et al., 2019a,b.

above sentence-level structural language. However, vocabulary and basic grammar (receptive and expressive) appear to be unaffected. Thus, the evidence reviewed suggests that measures of vocabulary and basic grammar cannot rule out higher-level language difficulty.

Further investigations are required to validate existing findings in a wider group, across different age ranges and with different IQ and autism severity. Other measures could also be explored with a particular focus on discourse analysis, spontaneously produced syntax and following instructions.

Perhaps surprisingly given these sex/gender differences in higher-level language abilities, questionnaire and interview data suggest that autistic females experience their language and communication difficulties similarly to autistic males, both in degree and type of impact reported. The parity of respondent accounts suggests that questionnaire and interview data may not be the best method for investigating sex/gender differences. The lack of observable differences when using these methods may reflect societal factors, with females and their parents naturally comparing their performance against the higher demands set by typically developing female groups. However, qualitative methods remain a critical tool for demonstrating the experience of the individual in both research and clinical domains.

Overall, then, it appears that the subtle language and communication difficulties outlined here may contribute to impact on functionality, social-interrelations and emotional well-being. These early findings should be consolidated with further empirical research. The relationship between subtle difficulties and emotional well-being is an area of particular concern due to the prevalence of mental health difficulties for this group.

Clinical Implications

This paper supports the notion of a specific female autism phenotype and extends this to the domain of language and communication differences. Awareness of this presentation is essential for accurate identification and diagnosis of autistic females without intellectual disability.

The presentation of subtle language and communication difficulties, in particular above sentence-level language, pragmatics (inference and discourse) and semantics, should be assessed in clinical settings. This should include direct assessment, observations and facilitated self-report. Basic structural language measures of vocabulary and sentence-level grammar should not be used to rule out communication difficulties.

Results from appropriate assessments of need should be used to guide targeted interventions. This should include managing the negative impact of language and communication difficulties on functionality, social-interrelations and emotional well-being.

Limitations

The literature in this area is sparse. It is also typified by smaller studies, and due to the wide range of measures, used overarching assumptions cannot be made with any certainty. In addition, many of the studies discussed are by necessity preliminary and exploratory. While these limitations mean that any conclusions drawn from the current paper must remain tentative, in itself this issue highlights an important point: linguistic profiles in the female autism phenotype are currently extremely poorly understood, and these gaps in our understanding may contribute to problems of mis- or under-diagnosis in this group. The current paper therefore highlights important avenues for future empirical work in this under-researched area.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/**Supplementary Material**, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by South West—Central Bristol Research Ethics Committee (November 2015). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

AS devised the research questions and was the lead researcher on the series of studies, which are discussed in this article. They were undertaken as part of her Ph.D., during which time she was supervised by JF and CA. All authors contributed to the development of methodology and data analysis across these studies. AS wrote this manuscript in consultation with this team of authors.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2021.621742/full#supplementary-material

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