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Speech, language, and literacy development in individuals with Down syndrome

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Speech, language, and literacy development in individuals with Down syndrome

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Editorial: Speech, language, and literacy development in individuals with Down syndrome

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Editorial on the Research Topic
Speech, language, and literacy development in individuals with Down syndrome

The ease and speed with which many children develop spoken language belies the complex processes that underpin it. Further, speech and language are fundamentally important for learning, thinking, reasoning, and remembering, as well as for communicating and fully participating in the social world. Mastering a written language is equally important and inextricably linked to spoken language, with each influencing progress in the other (Ellis and Large, 1988). Speech and language difficulties are common, and often persistent and severe, for individuals with Down syndrome, potentially impacting all aspects of cognitive and social development. This Research Topic contributes to our understanding of speech, language, and literacy development in individuals with Down syndrome and of effective interventions for this population.

Speech delays and difficulties are well-documented in individuals with Down syndrome, but there has been a lack of intervention work in this area. This may be linked to the field's limited understanding of underlying speech processes and their associated challenges. This is addressed in Madhaven et al. who propose a framework incorporating the biophysiological and environmental constraints to speech development and the interaction between them. This framework has implications for planning interventions for speech, language, and literacy. For example, they highlight the importance of somatosensory feedback children receive as they begin to produce sounds, which may be reduced in infants with Down syndrome.

As children with Down syndrome develop spoken language, many also develop dysfluencies, including stuttering. Although this is well-known in the field, there has been limited published research on this topic. In a review of the current literature, Hokstad and Næss identify that stuttering affects about 1 in 5 school age and adult individuals with Down syndrome and twice as many males as females but is less commonly diagnosed in pre-schoolers with Down syndrome relative to typically developing peers. They also note limitations of existing research and call for longitudinal studies to examine changes over time.

Turning to papers on language development, Mattie and Fanta explore the role of joint engagement in the vocabulary development of infants from 12 to 30 months. They report a perhaps unexpected finding that infants with more advanced joint engagement skills had fewer spoken words when measured concurrently and 9 months later. However, those with more advanced joint engagement skills at the later time point had higher receptive and expressive language scores. The authors suggest that at the earlier point in development, children may be using more advanced joint engagement behaviors to compensate for delayed spoken language and call for further research to unravel what is happening.

Two papers addressing early language development highlight the important role of shared book reading. Dulin et al. explore the home literacy environment (HLE) with a group of 11–14-month-olds using both a questionnaire and by recording and coding a parent-child shared book reading activity in the home. Richness of the HLE and quality of shared book reading activity, along with richness of the home language environment more broadly and the child's engagement in shared book reading, predicted receptive vocabulary 6 months later. Jeremic et al. review the literature on shared book reading as a language intervention for children with Down syndrome from birth to 6 years. They conclude that, despite limitations of existing studies, shared book reading can enhance children's language and communication. They identify that parents adapt their language for their child and that shared book reading provides opportunities for developing language. However, they also point out that it is possible that parents who are better at engaging their child in book reading may also provide higher quality language learning environments for their children throughout the day. This possibility should be considered in future longitudinal studies.

Romski et al. compare two interventions to teach first words to 24–29-month-olds over 12 weeks. Parents were coached in interventions embedded in natural play, in the clinic, and at home. One intervention focuses on teaching in a speech only format; the other includes the use of a speech generating device (SGD). The SGD group had more spoken words and intelligible utterances at the end of the intervention. The reasons for this warrant further study but may be the result of the SGD intervention providing more spoken repetitions of the target words.

Two studies look at later language development. Witecy et al. report on grammatical development from 4 to 17 years of age. Mastering verb agreement seems to be a prerequisite for mastering Wh- questions in German. They also report slower grammar learning from 10 years, suggesting there may be a critical period in which to develop grammar before this age. However, there are several possible explanations to be explored before reaching this conclusion. Children learn to talk so that they can communicate and may develop enough language to communicate effectively in their everyday world without needing to master grammar. More information about language learning environments and therapy experiences are also needed to inform understanding. The paper from Neitzel examines the narrative abilities of 10–20-year-olds. Patterns of individual differences in narrative responses suggest that those with more delayed non-verbal cognition and language abilities had more limited narrative abilities. There is a need to explore the narrative and communication abilities of young people in their everyday lives as well as experimental situations as these may differ.

Costanzo et al. report an intervention where Italian children and adults (5–29 years) were taught to communicate using an app which recognizes unclear speech and translates it into clear words. They report high levels of user satisfaction and some improvements in language abilities. A revolution in the way that technology may support this population may be seen in the near future, and this and the Romski et al. papers in this edition provide different examples of this.

Education and therapy services play a critical role in the progress and support of children and families, but the recent COVID pandemic brought these services to a halt. Pagnamenta et al. report the impact of this on families with children from 2 to 20 years. Parents reported that it was a stressful time, with some reporting a decline in their children's language and communication. What is learned from studies of the impact of COVID can be used to inform responses in future pandemics, but importantly, has also led to the positive development of tele-practice delivery of services.

On the assessment side, the paper from Channell et al. evaluates the validity of the Social Skill Improvement System Rating Scales for 124 individuals with Down syndrome from 6 to 17 years and demonstrates its validity and relations with other commonly used measures of behavior, autism traits, and executive functioning. The paper also highlights the relation between better expressive language abilities and social participation, and conversely, the link between poor expressive language and increased behavior difficulties. This highlights the role of language in all aspects of development, including behavior regulation and effective communication.

Moving into literacy, papers on early and adult readers include a study from Arango et al. which reports on the reading abilities and component skills of 6–10-year-old Chilean children learning to read Spanish. Measures included vocabulary, phonological awareness (PA), letter knowledge, sight word reading and verbal reasoning. Most measures showed improvement with age, but there was evidence of greater difficulty with PA and letter knowledge. This finding is consistent with studies of readers in the English language. Vocabulary was a predictor of word reading and some PA measures, which highlights the links between spoken language and reading. Importantly though, the authors point out that longitudinal studies are needed to explore these relations further.

Frizelle et al. report on co-construction of a reading assessment with 46 self-advocates in Ireland, examining what is considered relevant reading for them and constructing an accessible assessment. This is a pioneering example of inclusive research, involving the stakeholders from the outset and identifying what is relevant in their lives.

Not only does this Research Topic highlight the most recent research in speech, language, and literacy focused on individuals with Down syndrome, it also sets the stage for more impactful future research. As we see in this Research Topic, the field is moving beyond describing the communication difficulties of individuals with Down syndrome and into more research focused on interventions, meaningful and accurate assessments that do not demonstrate floor effects, community participation, and increasing diversity and the use of technology.

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Study of a set of reading precursors among Chilean children with Down syndrome

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Learning to read for children with Down syndrome is relevant because of the impact this ability has on learning and the development of autonomy. Previous research has described reading development in this population, but it is not clear if the process and precursors are the same in a transparent language like Spanish. This study explores performance in a set of precursors (phonological awareness, visual recognition, vocabulary, letter knowledge and verbal reasoning) in 42 children with Down syndrome between 6:0 and 10:11years. We hypothesized that the participants would have a lower performance than previously reported with children with typical development, particularly in tasks of phonological awareness, because the method for reading instruction in Chile with this population is usually the global method. Our results show that the precursors improve with age, that there are differences in performance between the skills assessed, and the ceiling effect was not observed as would be expected for children with typical development for the abilities assessed at these ages, which suggests that in the children assessed the precursors are not consolidated at these ages. These results suggest that the stimulation of phonological awareness and other reading precursors in children with Down syndrome is important for reading development.

KEYWORDS

reading, reading precursors, Spanish, Down syndrome, phonological awareness

Introduction

Reading is a necessary skill in the modern world, since it provides access to learning, facilitates the development of functional skills and drives social participation (Cologon, 2013). There are various theories about how reading ability development occurs, and although none is accepted as the only one, there are various common factors among them. In most theories it is proposed that reading depends on mental and linguistic sub-skills called precursors, which shape the system as a gestalt (Coltheart, 2006). The evidence for precursors' prediction of reading skill is incontrovertible (Adams, 1990), and it explains reading ability variations during typical development (National Reading Panel (US), National Institute of Child Health and Human Development (US), 2000; Torgerson et al., 2006). Some of the relevant reading precursors according to the literature are phonological awareness, decoding, visual recognition of words, language comprehension, and vocabulary (Scarborough, 2002). Studies among Spanish-speaking children show that these precursors are consolidated around the end of preschool education, between the ages of 4 and 6 years (Herrera and Defior, 2005; Vieiro-Iglesias et al., 2015; Morales et al., 2020). Therefore, formal reading instruction begins with these precursors already being consolidated, and by the end of second grade children are already expected to be competent readers.

Unlike the knowledge gained about reading development in typical development, there is little evidence about reading precursors' development in the presence of Down Syndrome, a congenital condition with high likelihood for intellectual disability (Sherman et al., 2007; de Graaf et al., 2016). The gap increases when considering native Spanish speakers.

Spanish is a Romance language and is the second most widely spoken language in the world (Eberhard et al., 2022). It is composed of 29 graphemes: the 26 letters of the English alphabet plus the letter ñ and the digraphs ch and ll (Goikoetxea, 2006). At the phonetic level there are 26 phonemes, of which 5 are vowels and the rest consonants. A syllable in Spanish has a maximum of 5 phonemes, with a maximum of two initial consonants (Defior and Serrano, 2014). The most frequent syllable structure is CV followed by CVC. Spanish has an alphabetic phonological orthographic system, and although there are geographical phonological differences, they do not generate comprehension problems between speakers (Defior and Serrano, 2014). The correspondence between graphemes and phonemes is almost biunivocal. Therefore, Spanish is considered a language with a transparent orthographic system due to the high grapheme-phoneme correspondence (Bravo-Valdivieso and Escobar, 2014). This has practical implications for learning to read. For example, children learn to read faster than in opaque orthographic systems (Seymour et al., 2003), preferably through syllabic-phonological methods. Reading is explicitly taught in the first year of basic education, around age 6, and decoding is typically consolidated during the second year of instruction (Defior and Serrano, 2014). Additionally, their orthographic transparency qualities impact the role and relevance of precursor skills. For example, although phonological awareness is relevant and emerges around the age of 4 (Acevedo, 1993), in the initial stages of reading formation it yields its predictive power in favor of variables more related to reading fluency, such as rapid automatized naming (Defior, 2008). It is also the case that, in Spanish as in other shallows orthographies, students acquire orthographic knowledge earlier, which they use to read words (Share, 1995; Goikoetxea, 2006).

It is important to note that education in Chile is compulsory from the first year of basic education, which children enter at age 6, to the last year of secondary education. Regarding preschool education, data from the Ministry of Education indicate that approximately 50% of Chilean children between the ages of 4 and 5 attend kindergarten, including children with disabilities (Subsecretaría de Educación Parvularia, G. de C, 2021). As in other countries, formal reading instruction begins in first grade, although the reading precursors are worked on in the preschool years (Subsecretaría de Educación Parvularia, G. de C, 2018). It is important to note that Chile has a unified national curriculum that all schools must follow regardless of whether they are regular or special schools, so every child in the country receives the same amount of reading instruction.

The present study intends to provide knowledge *via* analyzing the development traits of a set of reading precursors among a group of Chilean children with Down Syndrome. We follow the model of Scarborough (2002) who stated that competent reading has fluid and coordinated integration of two dimensions, namely recognition of written words and language comprehension. Within the recognition of written words, we can include phonological awareness, decoding and the visual recognition of words. Language comprehension includes conceptual knowledge as well as knowledge about linguistic structures, vocabulary, verbal reasoning, comprehension of figurative language, literary genres and their characteristics (Scarborough, 2002).

Considering that there is agreement about how the central reading precursors are phonological awareness, decoding, visual recognition of words, vocabulary and verbal reasoning, the study is centered on exploring these dimensions (National Reading Panel (US), National Institute of Child Health and Human Development (US), 2000).

Phonological awareness

This is a metalinguistic skill allowing its users to perceive and manipulate language units, and is indispensable for acquiring the alphabetical principle, that is, the notion that in alphabetic spellings such as Spanish, the written symbols represent sounds, and not other units such as ideas or words (Bravo Valdivieso, 2002). There is evidence that without this skill, it is not possible to move into more complex areas of learning to read such as inferential comprehension (Bianco et al., 2012), which is similar in languages with different levels of spelling transparency (Caravolas et al., 2005).

Various tasks have been used to evaluate phonological awareness, with the most common ones being segmentation of syllables and phonemes, rhyme recognition, and initial and final sounds and syllables (Goswami and Bryant, 2016). There is evidence suggesting that in the presence of typical development, Spanish speakers can achieve competence in syllable segmentation between the ages of 4 and 5 years (Vieiro-Iglesias et al., 2015; Morales et al., 2020) and for syllable discrimination between ages 5 and 6 years (Aguilar Villagrán et al., 2011).

Given the recognized importance of phonological awareness as a reading precursor, it was ultimately incorporated as a relevant variable to understand reading development in Down Syndrome. Pioneering studies marked the area, particularly the work of Cossu and Marshall (1990) who stated that efficient oral reading in fact did not necessarily require a good level of phonological awareness. This affirmation assumed, among other things, the idea that children with Down syndrome learned to read by methods which were qualitatively different from children with typical development. Their evidence came from observing children with Down syndrome who achieved oral reading, but had very low scores in phonological awareness tests, which presented a disassociation of domains as something peculiar to their genetic condition.

Later studies (Cupples and Iacono, 2000; Fletcher and Buckley, 2002; van Bysterveldt and Gillon, 2014) have debated this finding by showing that the exploration of phonological awareness in Down syndrome faces a sizable challenge, namely the adaptation of paradigms to get the participants to understand complex instructions. Therefore, more than being a group which cannot achieve phonological awareness, it is a problem of access to reliable information due to shortcomings in the measuring instruments. With adaptations to the experimental paradigms, it has been shown that children with Down syndrome substantially improve their performance on phonological awareness tasks over time and with systematic school instruction (Kay-Raining Bird et al., 2000), although it is not yet clear at what age this achievement can be expected. Predictive relations have also been documented between phonological awareness and working memory with reading performance in Down syndrome (van Bysterveldt and Gillon, 2014).

Research has shown that in people with Down syndrome, phonological awareness skills are compromised at different levels depending on the complexity of the task. In this sense, complex skills such as blending and segmenting sounds present low performance (Lemons and Fuchs, 2010a), while performance improves in basic tasks such as initial phoneme identification, although comparatively still below the performance of peers with typical development (Kalaycı and Diken, 2022). Rhyme detection tasks have also been found to be challenging for this population (Verucci et al., 2006; Næss, 2016), although there are also studies reporting rather moderate compromises (Kalaycı and Diken, 2022). Regarding syllable segmentation, the results are not conclusive, but with a tendency for these skills to be compromised (Verucci et al., 2006). It is reported that within the more complex phonological skills, children with Down syndrome present more difficulties with syllable segmentation compared to the blending tasks (van Bysterveldt and Gillon, 2014; Kalaycı and Diken, 2022). For other authors, however, the trade-offs are rather moderate. For example, Acarlar et al. (2002) in a sample of Turkish children, report that syllable segmentation is the easiest phonological task for this group. They mention that it is possible that the orthographic qualities of Turkish may explain this effect since Turkish words have short syllables, something similar with Spanish (Defior and Serrano, 2014). Results of the studies of the phonological strengths and weaknesses of people with Down syndrome are relevant because they provide clear perspectives for the reading interventions. For example, they suggest the need for a clear knowledge about the individual's phonological abilities in order to provide the necessary supports (Lemons, 2015; Hessling Prahl et al., 2022), which in turn impacts on word reading skills (van Bysterveldt and Gillon, 2014).

Letter knowledge

This consists of applying knowledge about the relations between letters and their sounds to pronounce the words and assign them meaning, and is a precursor of decoding (Beck and Juel, 1995). Visual recognition of words includes synthesizing phonemes or syllables to read larger units and with an associated meaning. Evaluation of letter knowledge is usually done *via* exploring knowledge of graphemes or by knowledge of letters' names.

Identifying letter sounds and names has been consistently acknowledged as an important predictor of reading (Adams, 1990; Whitehurst and Lonigan, 1998). Letter-sound knowledge facilitates the acquisition of the alphabetic principle, which in turn, helps children decode. Many studies also support the importance of explicitly teaching letter-sound correspondence to facilitate reading development (Ehri et al., 2001; Dehaene and Cohen, 2011; Solheim et al., 2018).

Evidence of interventions to enhance letter identification skills among children with Down Syndrome have yielded significant improvement. For example, bringing children's attention to printed letter features and phonemes during shared reading facilitated letter knowledge (Van Bysterveldt et al., 2007). Just like typically developing readers, children with Down Syndrome benefit from explicit, systematic, and supplemental instruction on code-related skills such as letter identification (King, 2020; King et al., 2022).

Some authors have described decoding as a relative weakness in children with Down Syndrome, as opposed to visual recognition of words (Verucci et al., 2006; Hulme et al., 2012), while other researchers have not found differences in these skills between children with typical development and Down syndrome on non-timed tasks. This would indicate that the problems lie in processing speed rather than in decoding (Snowling et al., 2002). It should be mentioned that decoding skills' development among children with Down syndrome has seen little study, which some authors associate with the difficulties of evaluation with tasks designed for children with typical development (Næss et al., 2012).

Visual recognition of words

One of the most influential models for explaining word reading is the double-route model (Coltheart, 2006). This model states that words can be read by a phonological pathway, when they are new or infrequent words, or else by a visual or lexical route, which is used for reading familiar or frequent words. Readers with more reading experience achieve quicker visual recognition of words and automate this process to consolidate it, when they are native Spanish speakers, around age 6.

Visual recognition of words is a skill which helps free up cognitive resources originally oriented towards decoding words, which can now be redirected towards processes related to reading comprehension (Mimeau et al., 2018). Among people with Down Syndrome, it has been found that word reading is their main reading strength, leading to a focus on intentional learning of word reading from global methods (Troncoso and Flórez, 2011). The studies show that the reading profile of people with Down syndrome presents a discrepancy between word reading level and reading comprehension (Næss et al., 2012), compared with the profile of reading comprehension problems in a typical population. However, it is possible that this profile is not present in a Latin American context, where research has shown that children with Down syndrome have difficulties both in word reading skills and reading comprehension (Hernández Salazar and Talou, 2005).

Vocabulary

Vocabulary is one of the best predictors of reading comprehension once decoding processes have become automatic. In fact, vocabulary and comprehension have a bidirectional relation that increases over time (Torgesen et al., 1997; Tannenbaum et al., 2009; Quinn et al., 2015). Children's vocabulary develops through social verbal interactions prior to learning how to read, and the amount of vocabulary knowledge is strongly associated with socioeconomic background (Hoff, 2003). Children's vocabulary in the early years accounts for a significant amount of variance in reading comprehension, both from a linguistic and cognitive perspective (Duff et al., 2015).

The bidirectional relation between vocabulary knowledge and comprehension can be explained by the fact that understanding the meaning of a word allows a reader to better understand a passage and, as readers become better comprehenders, their vocabulary also increases. At the same time, one can infer that vocabulary and comprehension also contribute to conceptual knowledge, and therefore are strongly correlated (Rupley, 2012). This relation can also be explained if vocabulary and comprehension are part of what is defined as verbal aptitude, so that students who know more about words and concepts are better at building meaning from text (Anderson and Freebody, 1981). From a pedagogical perspective, the bidirectional relation is likely to have more implications for the development of both comprehension and vocabulary.

Studies of the relation between vocabulary and reading have received attention for both children with typical development and with

Down syndrome (Kay-Raining Bird et al., 2000) and are often studied *via* exploring receptive vocabulary. Evidence suggests that this skill is consolidated around ages 4 or 5 in children with typical development (Cáceres Zuñiga, 2018).

Although most studies in Down syndrome have been done with Anglophone populations, a significant advance has arisen from knowledge about vocabulary in this group following the Spanish standardization of the MacArthur-Bates Communicative Development Inventories (Mariscal et al., 2007; Galeote et al., 2011; Checa et al., 2016).

Regarding the relation between vocabulary and reading abilities in children with Down syndrome, previous studies have shown strong correlations between these abilities, which tend to be higher in the children with Down syndrome, when compared to children with typical development. For example, Nash and Heath (2011) found stronger correlations between vocabulary and reading comprehension in children with Down syndrome than in those with typical development. Also, in a longitudinal study, Hulme et al. (2012) found that vocabulary was a strong predictor of reading performance both for children with Down syndrome and in children with typical development, but it was a stronger predictor for the former than for the latter.

Verbal reasoning

This is the capacity to process and use orally presented information and properly integrate it to understand its meaning and manipulate verbal information (Language and Reading Research Consortium, 2017). The influence of verbal abilities such as oral comprehension and verbal working memory on reading development is documented among children with typical development (Lervåg et al., 2018), but there are few studies in Down syndrome (Boudreau, 2002; Roch et al., 2015).

Oral comprehension level tends to be considered a good indicator of verbal reasoning, achieving a good performance level between 4 and 5 years (Florit et al., 2009). Prior studies have shown that this dimension is a weakness for people with Down Syndrome, characterized by difficulties in production and syntactic comprehension (Boudreau and Chapman, 2000), but there are no available studies exploring at which age one can expect performance relatively similar to that observed in typical development when reaching a competent level.

In general terms, there is little literature exploring reading precursors in children with Down syndrome who are native Spanish speakers. There are some studies which have shown the extended belief that for children with Down syndrome reading is equivalent to decoding, without incorporating other predictive dimensions of comprehension, which is not supported by available evidence (Cupples and Iacono, 2002; Jiménez and Flórez-Romero, 2013; de la Cruz Paulino, 2017).

The incorporation of the global method – which emphasizes the reading of words as a Gestalt and not the teaching of the sub-lexical units of language such as sounds and morphemes – dominated methods for teaching reading to children with intellectual disabilities in general, and Down syndrome in particular, for over 30 years (Troncoso and Flórez, 2011). Particularly in Chile, this is still the officially recommended method to teach reading to this population group (Ministerio de Educación, 2008) and is thus the method adopted across the board in special schools and civil society foundations which offer alternative accompaniment to the population.

However, studies have shown that systematic and explicit teaching of the association between letters and their sounds is related with improvements in this population (Lemons and Fuchs, 2010b), meaning that the global method is insufficient (Cologon et al., 2011). Studies of reading skill development require an exploration which responds to the complexity of the process and which, in the case of children with Down syndrome who tend to need help, goes beyond decoding, and allows us to understand the strong and weak points of the process for more efficient support and accompaniment.

This exploratory study was aimed to analyze a set of reading precursors in children with Down syndrome, with a hypothesis that differentiated development levels will be observed compared to those previously reported in the literature about typical development for Spanish speaking children in these precursors, with phonological awareness being the dimension where higher differences are expected, given the lack of attention which it receives under the reigning instructional model for children with Down syndrome within the country, the global teaching method.

Methods

This study follows a within-subject design, based on the analysis of cross-sectional measures of performance on tasks exploring reading precursors among a group of school-age Chilean children.

Participants

The study included 42 Chilean children with Down syndrome (21 girls and 21 boys) between 6:0 and 10:11 years (*M* = *7.98, SD* = *1.44*). Twenty-three participants attended special schools and 19 attended regular schools. The entire sample received educational and learning accompaniment outside of school hours *via* special institutions to support people with Down syndrome. All the schools of the participants stated that they use the global method as the main reading instruction method, and that they follow the indications of the national curriculum. For the analysis, the sample was divided into three age groups: 6:0–7:11 years (*n* = 17), 8:0–9:11 years (*n* = 17) and 10:0–10:11 years (*n* = 8). We decided to make this division by age groups since reading development is related to both age and grade level, and these are the ages that typically correspond to grades 2, 3, and 4 in Chile.

Inclusion criteria were: (1) having confirmed Down syndrome with a karyotype, (2) receiving reading instruction with the global method, (3) presenting appropriate functioning in sensory systems or adequate correction with glasses or hearing aids, (4) having authorization from their legal guardians by signing informed consent and (5) being able to provide consent with a witness for their participation. The exclusion criteria were: (1) having an uncorrected sensory difficulty, and (2) having undergone any type of surgical intervention and/or hospitalization during the month prior to the reading skill evaluation.

Instruments

We adopted a strategy of evaluation which has been applied in prior studies done with children with Down syndrome who were native English speakers, where paradigms had to be adapted (e.g., van Bysterveldt and Gillon, 2014). Experimental evaluation tests were applied for reading precursors whose validity is documented in previously published studies. This decision is due to the lack of packaged exploration instruments for reading precursors with proper psychometric evidence for a Spanish-speaking Down syndrome population. While there is an extant Spanish version of the MacArthur-Bates Communicative Development Inventories which has been tested on children with Down syndrome in Spain, and which explores vocabulary (Galeote et al., 2012), given the discrepancies between the Spanish used in Spain and Chile, it is not recommendable to use it in our context. Along with this, the age range which is the target for this test (8–30 months) is not the age we wanted to assess in the present study.

The evaluation of phonological awareness included syllable discrimination and segmentation tasks. For decoding, we considered grapheme-phoneme recognition tasks and letter name knowledge. We also used tasks to explore visual recognition, receptive vocabulary, and verbal reasoning. Table 1 presents task descriptions, their variable of interest, and examples of studies which have used the paradigm and offered validity and reliability evidence *via* internal consistency with Cronbach's Alpha.

Procedure

Procedures incorporated in this study were supervised by the Ethics Committee at the hosting University.

Children with Down syndrome were invited *via* six support organizations located within the urban area of Santiago, Chile. Parents signed an informed consent authorizing their child's participation, and the children themselves provided verbal assent.

Evaluations were done by a research assistant with postgraduate neuropsychology training. Tests were applied following a standardized protocol, always in the same order, with an average time of 30 min. The children were evaluated in the organizations they attend or in appropriate spaces at the University. They were accompanied by their parents in all cases and were able to ask to stop or receive help whenever they required. Qualitative performance reports were delivered to each family, accompanied by recommendations for stimulating reading development and a users' manual for La Mesita, a tablet-based game designed to promote reading development in children with intellectual disability (Tenorio, 2016).

Data analysis

Data analysis was done considering three precursors associated with visual word recognition according to the model from Scarborough (2002): phonological awareness, decoding and visual recognition. We also analyzed two precursors associated with language comprehension under this model: vocabulary and verbal reasoning.

To supply reliability and validity evidence for the experimental tests, we followed recommendations from the international measurement quality standards for psychology and education (American Educational Research Association, American Psychological Association and National Council on Measurement in Education, 2014). This offered reliability evidence analysis *via* internal structure exploration using Cronbach's Alpha. Validity evidence is based on content study and paradigm implementation with broad prior use.

We present descriptive data with the means and standard deviations from the sample and consider an analysis of simple correlations with age as a variable, and bivariate Pearson correlations controlled by participants' age in years. To analyze achievement differences between precursors, we compared performance medians for each age group using one-way ANOVAs for the variables of interest. For each comparison we tested the homoscedasticity with Bartlett's test. Post-hoc comparisons with Holm-adjusted p-values were used to study the difference points.

All analyses were performed in R 4.2.1 (R Core Team, 2022) with packages stats (R Core Team, 2022), psych (Revelle, 2020) and tidyverse (Wickham et al., 2019).

Results

Preliminary data inspection shows variance, without any floor effect, a commonly observed effect when evaluating children with Down syndrome (Kennedy and Flynn, 2003). For all variables we observe a trend towards improved performance as ages rose (Table 2).

The simple correlations matrix shows a positive, strong, and significant correlation between age of the participant in years and vocabulary, oral comprehension, final syllable discrimination, word recognition and syllable segmentation. No correlation was found for age and grapheme recognition, knowledge of letters' names and syllable auditory discrimination (Table 3).

When analyzing the partial correlations matrix (Table 4), there is a notable presence of positive, strong, and significant relations between vocabulary and the other variables explored, apart from oral comprehension and syllable discrimination. There are also positive, strong, and significant correlations between word recognition and syllable segmentation, grapheme recognition, vocabulary, and knowledge of letter's names. There is another similar pattern in the grapheme recognition and knowledge of letter's names variables.

In the comparison between age groups there are several statistically significant differences in favor of older children with small effect sizes (Cohen, 1992; Dunst and Hamby, 2012; Bakker et al., 2019). Regarding phonological awareness exploration, there are significant differences by age group for final syllable discrimination ($F = 3.41$, $p < 0.05$, $\eta^2 = 0.166$) and syllable segmentation ($F = 10.22$, $p < 0.001$, $\eta^2 = 0.258$) but not syllable auditory discrimination ($F = 1.44$, $p = 0.269$, $\eta^2 = 0.074$). In letter knowledge, while there is a statistically significant difference for grapheme recognition ($F = 5.23$, $p < 0.01$, $\eta^2 = 0.230$), there is no such difference in knowledge of letter names ($F = 1.45$, $p = 0.249$, $\eta^2 = 0.076$). In visual word recognition, there are significant differences in word reading ($F = 6.16$, $p < 0.05$, $\eta^2 = 0.350$). There are also significant differences for vocabulary ($F = 8.23$, $p < 0.001$, $\eta^2 = 0.320$) and oral comprehension ($F = 5.41$, $p < 0.01$, $\eta^2 = 0.242$).

The post-hoc tests show that all variables appear to stabilize between the second age group (age 8 to 9) and the third one (10 years), as no significant differences are found between these two older groups (Table 5). Regarding the comparisons between the first age group (6 and 7 years) to the second one (age 8 and 9) and between the first and the third one (age 10) there are three patterns: a significant change between the two younger groups that stabilizes afterwards (Grapheme recognition: $F = 4.027$, $p < 0.01$), a significant change when comparing the youngest and oldest groups (Final syllable discrimination: $F = 5.051$, $p < 0.05$), and significant differences between both the two younger groups and the youngest and oldest groups (Syllable segmentation: $F = 7.161$, $p < 0.05$ and $F = 17.075$, $p < 0.01$; Word recognition: $F = 11.876$, $p < 0.01$ and $F = 11.460$, $p < 0.01$; Vocabulary: $F = 13.380$, $p < 0.001$ and $F = 6.998$, $p < 0.05$; and Oral comprehension:

TABLE 1 Description of experimental paradigms.

General dimension	Subdimension	Paradigm	Description	Reference studies	Cronbach's alpha
Written word recognition					
Phonological awareness		Syllable auditory discrimination	The child repeatedly listens to a syllable, e.g., /do/, and must touch the screen when they hear a different syllable, e.g., /ma/. Items are calibrated in a progressively more complex sequence determined by syllables' phonetic similarity.	Rosas et al. (2011, 2017); Rosas (2013); Abello et al. (2014); Cadavid-Ruiz et al. (2016)	0.694
		Final syllable discrimination	Three images of objects are presented. The child hears the name of each object presented and chooses the one with a different final syllable. Information presentation is auditory, without any input from written words.	Rosas (2013); Abello et al. (2014); Escobar and Meneses (2014); Cadavid-Ruiz et al. (2016); Rosas et al. (2017)	0.722
		Syllable segmentation	The child hears a word and must segment it into syllables *via* a drum which is used by touching a screen. The items were calibrated for progressively rising difficulty according to the words' extension and syllabic structure.	Rosas (2013); Cadavid-Ruiz et al. (2016)	0.782
Letter knowledge		Grapheme recognition	A letter graph is presented, and the child must choose the representation for the sound of graph they see between four alternatives. The test is calibrated in progressively rising difficulty determined by the order in which letters are taught in a Chilean school context: first vowels, then consonants.	Rosas (2013); Cadavid-Ruiz et al. (2016)	0.768
		Knowledge of letters' names	The child hears the name of a letter and must pick the right choice from four possible graphs. We used the same items from the grapheme recognition task.	Ricci (2011); Rosas (2013); Cadavid-Ruiz et al. (2016); Rosas et al. (2017)	0.791
Visual recognition		Word Reading	The participant must read increasingly complex words out loud, as they appear on screen. Complexity is determined by the words' length and syllabic structure. Reading precision is scored.	Rosas et al. (2011); Rosas (2013)	0.897
Language comprehension					
Vocabulary		Vocabulary	Four images are presented, and the evaluator says a word out loud. The child must indicate which image corresponds to the word they heard. For instance, the evaluator says "dog" and the child must choose between a dog, a cat, a pear, and a moon. The test is calibrated for increasing complexity determined by words' frequency and length. The key variable is precision.	Dunn and Dunn (1997); Strasser et al. (2010)	0.843

(Continued)

TABLE 1 (Continued)

General dimension	Subdimension	Paradigm	Description	Reference studies	Cronbach's alpha
Verbal reasoning		Oral Comprehension	The evaluator says a phrase and the child must choose which of three images best represents it. For instance, the evaluator says: "The cat sleeps." Three images appear: a dog sleeping, a cat playing, and a cat sleeping. The key variable is precision.	Rosas et al. (2011); Rosas (2013); Cadavid-Ruiz et al. (2016)	0.675

TABLE 2 Descriptive statistics for reading precursors by age group.

		6:0–7:11 M(SD)	8:0–9:11 M(SD)	10:0–10:11 M(SD)
Recognition of written words				
Phonological awareness	SAD	0.39 (0.65)	1.15 (1.63)	0.88 (1.46)
	FSD	1.00 (1.30)	1.88 (1.09)	2.25 (1.17)
	SS	0.87 (0.92)	2.50 (2.25)	3.25 (1.49)
Letter Knowledge	GR	2.67 (1.23)	4.81 (2.11)	4.00 (2.27)
	KLN	3.07 (1.79)	4.25 (2.05)	4.13 (2.48)
	WR	2.57 (1.91)	4.43 (0.65)	4.57 (0.79)
Language comprehension				
Vocabulary	Voc	5.33 (2.47)	8.19 (1.76)	8,00 (1.93)
Verbal reasoning	OC	2.29 (1.38)	3.50 (1.27)	3.88 (0.84)

SAD= syllable auditory discrimination, max = 6; FSD = Final syllable discrimination, max = 6; SS = Syllable segmentation, max = 6; GR = Grapheme recognition, max = 7; KLN = knowledge of letters' names, max = 7; WR = Word reading, max = 12; Voc = Vocabulary, max = 10; OC = Oral comprehension, max = 5.

$F = 6.310$, $p < 0.05$ and $F = 8.650$, $p < 0.01$). No ceiling effect was observed in any variable.

Discussion

This study explored the performance of a group of school aged children with Down syndrome who received reading instruction with the global teaching method, on a set of reading precursors. We analyzed the variables which took part in emergent reading among this population, to reflect on the transformations which should be considered when designing educational programs oriented towards reading instruction for children with Down syndrome.

The first notable result is that none of the variables explored showed a ceiling effect for any of the three age groups which suggests that for the children in our sample, reading precursors are not yet consolidated at age 10. The evidence from previous studies shows that in children with typical development, all the precursors explored in our study are consolidated within the first age range explored in this study (6:0 to 7:11 years) (Rosas et al., 2011; Bravo-Valdivieso and Escobar, 2014; Escobar and Meneses, 2014). Considering the delay normally observed in Down syndrome, it should lie within the higher age range for this group (10:0–10:11 years). This finding is in line with our general hypothesis and provides evidence for a general delay of reading precursors within the evaluated group, indicating that children with Down syndrome need even more time to consolidate these skills and thereby, move forward in the formal process of learning to read.

In almost all the variables explored we also observed progressive improvement in performance, which shows that children with Down syndrome can learn these abilities, as previous studies have shown (Boudreau, 2002; Goetz et al., 2008; Baylis and Snowling, 2011). Although in this study we used experimental tasks and did not assess abilities through standardized tests to measure the variables evaluated, there is ample evidence about the ages at which Spanish-speaking children develop the reading precursors explored in this study (Seymour et al., 2003; Ziegler and Goswami, 2005; Míguez-Álvarez et al., 2022). This evidence allows us to suggest that the achievement level observed did not reach the same level that should be expected for children with typical development according to the literature, and also that it was not the same expected for children with Down syndrome who received instruction focused on stimulating phonological awareness, according to available reports (Baylis and Snowling, 2011).

One notable result from this study is the qualitative leap observed in performance quality at age 8 for variables where age progression was significant. We found significant differences between the first age group (ages 6 and 7) and the other two age groups in the syllable segmentation, word reading, vocabulary, and oral comprehension tasks. In the grapheme recognition task, there was a significant difference between the first and second (ages 8 and 9) age groups, and in the final syllable discrimination task a significant difference was found between the first and the older age groups. The differences found in the overall mean comparison analyses in these variables showed small effect sizes. In these variables our results suggest that children with Down syndrome have a significant performance improvement after age 7, according to the differences found between the three group ages compared in this study. It is complex to offer a conclusive explanation for this finding given the scope of this study, but we may hypothesize that this corresponds to the change documented in Spanish speaking children with typical development towards the end of their first year in primary school (Rosas et al., 2011; Escobar and Meneses, 2014), when reading instruction begins in schools nationwide, and which in turn drives the relevant transformation in the developmental trajectory of working memory, as previous studies with typically developing children have shown (Demoulin and Kolinsky, 2016). Various authors have suggested that working memory is a weakness in the Down syndrome cognitive performance profile, with an atypical trajectory whose achievements are more tardy (Brock and Jarrold, 2005; Lanfranchi et al., 2012). One might think that, as a function of the bidirectional relation between these two dimensions (Peng et al., 2018), the delayed appearance of these precursors within the evaluated group is related with the documented developmental delay of working memory.

TABLE 3 Simple correlations between variables of interest and age in years.

	SAD	FSD	SS	GR	KLN	WR	Voc	OC	Age
SAD	1	0.044	0.332*	0.281	0.244	0.084	0.222	0.169	0.121
FSD		1	0.23	0.304	0.375	0.478**	0.422	0.169	0.363*
SS			1	0.565**	0.533*	0.493*	0.593**	0.383	0.399**
GR				1	0.765***	0.567***	0.769***	0.550**	0.303
KLN					1	0.518***	0.664***	0.391	0.184
WR						1	0.802***	0.301	0.436**
Voc							1	0.426**	0.393*
OC								1	0.491**
Age									1

***$p < 0.001$; **$p < 0.01$; * $p < 0.05$. SAD = syllable auditory discrimination; FSD = Final syllable discrimination; SS = Syllable segmentation; GR = Grapheme recognition; KLN = knowledge of letters' names; WR = Word reading; Voc = Vocabulary; OC = Oral comprehension.

TABLE 4 Partial correlations between variables of interest controlled by age in years.

	SAD	FSD	SS	GR	KLN	WR	Voc	OC
SAD	1	0	0.312*	0.258	0.227	0.035	0.191	0.263
FSD		1	0.1	0.218	0.336	0.382*	0.326	−0.012
SS			1	0.508***	0.510***	0.387*	0.517***	0.235
GR				1	0.757***	0.507***	0.742***	0.483*
KLN					1	0.495***	0.654***	0.351
WR						1	0.762***	0.112
Voc							1	0.292
OC								1

*** $p < 0.001$; **$p < 0.01$; * $p < 0.05$. SAD = syllable auditory discrimination; FSD = Final syllable discrimination; SS = Syllable segmentation; GR = Grapheme recognition; KLN = knowledge of letters' names; WR = Word reading; Voc = Vocabulary; OC = Oral comprehension.

TABLE 5 *Post-hoc* analyses for the variables with significant differences by age group.

Variables / comparisons		Group 1 – Group 2		Group 1 – Group 3		Group 2 – Group 3	
		F	*p*	F	*p*	F	*p*
Phonological awareness	FSD	4.027	0.055*	5.051	0.036*	0.606	0.445
	SS	7.161	0.014*	17.075	0.002*	0.948	0.342
Letter knowledge	GR	11.778	0.002*	3.397	0.079	0.756	0.394
Visual recognition	WR	11.876	0.003*	11.460	0.003*	0.173	0.686
Vocabulary	Voc	13.880	0.001*	6.998	0.015*	0.057	0.814
Verbal reasoning	OC	6.310	0.018*	8.650	0.008*	0.571	0.458

Group 1: 6–7 years; Group 2: 8–9 years; Group 3: 10 years; SD = syllable discrimination; FSD = Final syllable discrimination; SS = Syllable segmentation; CG = Grapheme recognition; KLN = knowledge of letters' names; WR = Word reading; Voc = Vocabulary; OC = Oral comprehension.

We were also surprised to find no significant correlations among the three phonological awareness tasks assessed, which may seem counterintuitive given that the three tasks tap into the same underlying ability. Phonological awareness is a broad term that includes the ability to identify and manipulate sounds in language (Yopp and Yopp, 2009), but it entails tasks and levels of manipulation that vary in their complexity along a continuum. For example, identifying rhymes is easier than segmenting or blending syllables, and segmenting syllables is easier than segmenting sounds. This may explain why these tasks are not significantly correlated. On the other hand, our sample shows that children's ability to discriminate final syllables (FSD) and to segment syllables (SS) increased with age and these skills were significantly correlated with print-related skills such as grapheme recognition (GR) and letter name knowledge (KLN), which illustrates the contribution of phonological awareness to the acquisition of the alphabetic principle (Sulzby and Teale, 1991) that is necessary for word reading.

One element from the correlations matrix that stands out is the relations between vocabulary and the other dimensions explored. There is ample documentation of the role which vocabulary plays as a predictor for decoding and phonological awareness, for children with typical development and Down syndrome alike, among native English speakers (Lonigan, 2007; Van Bysterveldt et al., 2010). This finding makes sense when considering previously reported facts and suggests a similar pattern even in the presence of languages with different levels of orthographic transparency. Since it has been reported that vocabulary can be a causal variable for reading development among children with Down syndrome who are native English speakers (Carr, 2000; Laws and Gunn, 2002), future longitudinal studies should observe this relation in Spanish-speaking children.

Another relevant finding is the correlation between syllable segmentation, a skill particular to phonological awareness, and the variables explored in the recognition of the written word and language comprehension. This finding offers evidence favoring the hypothesis that phonological awareness is also important for reading development in children with Down syndrome (Fletcher and Buckley, 2002; Lemons and Fuchs, 2010b), although preliminary studies described its absence (Cossu et al., 1993; Evans, 1994). This result is particularly important in an educational context where it is necessary to review official recommendations to use global teaching methods for reading among children with Down syndrome.

This study has various limitations. First, it is an exploratory study with a sample which is considered to be a "good size" according to international parameters (Dunst and Hamby, 2012; Bunster, 2021), but which would doubtlessly benefit from expanding the number of participants so as to achieve greater analytical potency. Second, no measurements were taken for reading speed and reading pseudo-words, which are considered central skills for reading evaluation. Third, given the size of the analysis groups, it was not possible to perform regression analyses to establish the weight of the variables studied on some measures such as word reading. Fourth, given that it was a cross-sectional study it is not possible to establish a causal relation between the variables, and the trajectory analysis was incomplete. Future studies should have a longitudinal perspective in order to carry out predictive studies regarding reading development and better understand the developmental trajectories of reading in Down syndrome. Fifth, another limitation is that the phonological awareness tasks addressed only syllable discrimination and awareness, and not phoneme awareness. Future studies should include a more profound assessment of phonological awareness skills. Another relevant limitation is that the three groups are not equivalent as the older age group has fewer participants than the other two groups and includes only 10-year-olds, while the other two groups include children across 2 years of development. Finally, all measures in our study are experimental, which limits the extent to which our findings can be generalized. The inclusion of standardized measures of reading precursors and abilities in future research is necessary.

The results obtained allow us to suggest that the reading skills of a group of children with Down syndrome who are native Spanish speakers are sensitive to change and improve with age. The trend observed between age groups suggests that there is an underlying developmental trajectory similar to that described in children with typical development, an affirmation which should be contrasted in future studies. If similar projections are proved, and according to what previous studies have suggested (Lemons and Fuchs, 2010a,b; Baylis and Snowling, 2011; Burgoyne et al., 2012), it will be relevant to incorporate phonological awareness stimulation on the daily practice of teaching children with Down syndrome to read, as well as targeted instruction in other relevant reading precursors such as vocabulary and letter knowledge.

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 Comité de ética de Ciencias Sociales, Pontificia Universidad Católica de Chile. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

MT, PA, JE, and RR contributed to conception and design of the study. AA performed the statistical analysis. MT, PA, and JE wrote the first draft of the manuscript. AA and PO wrote sections of the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Using the Social Skills Improvement System (SSiS) Rating Scales to assess social skills in youth with Down syndrome

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Introduction and Methods: This study provides preliminary data on the Social Skills Improvement System (SSiS) Rating Scales Parent Form to measure social skills in a sample of 124 children and adolescents with Down syndrome (DS) ages 6–17 years.

Results: Overall, participants demonstrated relatively mild symptoms, with the sample's average standard score falling within 1 standard deviation from the mean of the normative sample for the social skills (M=92, SD=15) and problem behaviors (M=104, SD=12) domains (normative sample M=100, SD=15 for both domains). However, a wide range of scores was observed across the sample for the composite and subscale scores. Differential patterns were also observed by subscale. For some subscales (i.e., Cooperation, Assertion, Responsibility, Engagement, Externalizing, Hyperactivity/Inattention, and Autism Spectrum), a disproportionate number of participants scored in the below average (i.e., lower levels of social skills) or above average (i.e., more symptomatic in problem behaviors or autism spectrum) range relative to the normative sample; for other subscales (i.e., Communication, Empathy, Self-Control, Bullying, and Internalizing), participants' score distribution aligned more closely to that of the normative sample. SSiS composite scores correlated in the expected directions with standardized measures of autism characteristics, executive function, and expressive language.

Discussion: This study provides some of the first evidence validating the use of the SSiS in youth with DS, filling a gap in standardized measures of social functioning in this population.

KEYWORDS

social skills, down syndrome, challenging behavior, problem behaviors, social interaction, intellectual disability

1. Introduction

Individuals with Down syndrome (DS) show a distinct yet complex phenotype that affects the language, cognitive, and social skills they use to interact and communicate with others (Iarocci et al., 2008; Cebula et al., 2010; Grieco et al., 2015; Thurman and del Hoyo Soriano, 2021).

Research on social development in DS has focused mostly on early foundational skills, identifying several relative strengths (e.g., eye gaze, gestures, vocalizations, joint attention; Fidler, 2006; Fidler et al., 2008; Cebula et al., 2010; Thurman and del Hoyo Soriano, 2021). However, less is known about how individuals with DS use these foundational skills during social interactions in later childhood and adolescence. This lack of information represents a critical gap in the literature, given the range of individual differences in outcomes related to social development, such as independent living, employment, community participation, and quality of life, that have been reported among adults with DS (Iarocci et al., 2008; Scott et al., 2014; Jevne et al., 2022; Loveall et al., 2022). One major barrier is that few measures of social skills have been established in this population (Esbensen et al., 2017; Schworer et al., 2021). To address this barrier, the purpose of the current study was to evaluate the Social Skills Improvement System (SSiS) Rating Scales (Gresham and Elliott, 2008) for assessing social skills in a large sample of children and adolescents with DS.

Successful social interaction requires the coordination of many skills. For example, from the framework of social information processing theory (Crick and Dodge, 1994), an individual must show a general social orientation to pay attention to and encode the social cues around them. Then, the individual must use social cognition to interpret their social partner's verbal and nonverbal communication and engage in social reasoning to make internal evaluations, ultimately deciding on a behavioral response. These processes also require the integration of other skills related to attention, emotion understanding, language processing, and emotion regulation (Lemerise and Arsenio, 2000; Grazzani et al., 2018).

Current research on key social functioning skills in DS indicates that children and adolescents often show floor effects or low performances on measures of social cognition that involve tasks like perspective-taking, social reasoning, and social problem-solving (Abbeduto et al., 2008; Hahn et al., 2013; Ashby et al., 2017; Barisnikov and Lejeune, 2018; Martin et al., 2018; Schworer et al., 2021). However, these tasks tend to rely heavily on language processing and executive function, known areas of difficulty in DS, posing a challenge for assessing social cognition *per se* (see Channell and Loveall, 2021). Similarly, youth with DS show difficulties in aspects of pragmatic language, again using tasks that require higher-order social cognition (i.e., perspective-taking or theory of mind) and language (Lee et al., 2017; Smith et al., 2017). If individuals with DS demonstrate lower performances on these tasks in experimental settings, it is possible this translates to difficulties during day-to-day social interactions, especially as they progress into adolescence when social demands increase (Iarocci et al., 2008). However, it is also possible that in more naturalistic settings, individuals with DS may benefit from situational and nonverbal cues (e.g., gesturing, eye gaze, emotion recognition) to help them navigate social interactions. For this reason, more ecologically valid assessments are needed.

In clinical settings, a common approach for assessing social skills in childhood and adolescence is through informant report (e.g., caregiver, teacher, self, peers). Informant report measures are particularly helpful for understanding social behavior in naturalistic settings and across different environments (e.g., home, school, peer interactions). One such measure developed for this purpose is the SSiS (Gresham and Elliott, 2008), which is the newer version of the Social Skills Rating System (SSRS; Gresham and Elliott, 1990). The SSRS and SSiS were developed for clinical use to identify children who have delays in social skills and challenging behaviors that affect social interaction to develop intervention targets. To our knowledge, only two studies have examined the SSRS in small samples of 4- to 6-year-olds with DS through parent (Guralnick et al., 2009) and teacher (Guralnick et al., 2011) report. In both studies, young children with DS had lower social skills standard scores than both chronological and mental age-matched neurotypical peers, suggesting that even in early childhood, this measure is sensitive to delays in social skills in DS. These findings suggest that even in early childhood, children with DS may show a profile of difficulties in some areas of social development and in related domains that are also critical to social interaction [i.e., expressive language, internalizing and externalizing problems, hyperactivity, and distractibility (Guralnick et al., 2009, 2011)]. To our knowledge, no study to date has systematically measured social skills across older children and adolescents with DS. Such information is needed to understand social skills in DS within the framework of the social demands experienced by older youth.

A few studies have used other caregiver report measures of social challenges that were originally developed to capture autism symptoms [i.e., Social Responsiveness Scale (SRS; Constantino and Gruber, 2005); Social Responsiveness Scale-2nd edition (SRS-2; Constantino and Gruber, 2012); Children's Social Behavior Questionnaire (Hartman et al., 2007)] to examine social skills in youth with DS. Across these studies, children and adolescents with DS tend to score in the elevated range, showing more social challenges relative to chronological age- and sex-based norms from the general population (van Gameren-Oosterom et al., 2013; Channell et al., 2015; Channell, 2020; Schworer et al., 2021). Although these measures were designed to capture challenges in social communication and interaction in the context of autism risk, they show potential as measures of broader social outcomes for individuals with DS (Schworer et al., 2021). However, more research is needed to support their use in this population and to systematically examine profiles of social skills in youth with DS. The current study seeks to address the latter.

In addition to their clinical use, measures of social skills can contribute to the understanding of the DS phenotype. Over the past few decades, a robust body of research has characterized the behavioral phenotype associated with DS in different developmental domains (see Grieco et al., 2015; Thurman and del Hoyo Soriano, 2021 for reviews). For example, individuals with DS tend to show relative difficulties in the domains of expressive language (Abbeduto et al., 2007; McDuffie et al., 2017), motor development (Winders, 2013; Frank and Esbensen, 2015), and auditory processing (Conners et al., 2011), with relative strengths in aspects of visuospatial processing (Yang et al., 2014) and early social skill development (Fidler et al., 2008). This profile begins to emerge early in life and builds over time as children with DS adapt and develop strategies to interact with the world around them (Fidler, 2005; D'Souza and D'Souza, 2022). Because development in one domain affects subsequent development in related domains, unique intra-individual profiles continue to evolve across childhood (D'souza et al., 2017; Fidler et al., 2019; D'Souza and D'Souza, 2022). Furthermore, these developmental profiles affect, and are affected by, the individual's environment and neurogenetics, resulting in considerable inter-individual differences in developmental trajectories (Cebula et al., 2010; Karmiloff-Smith et al., 2016). By examining social skill profiles in children and adolescents with DS alongside related domains such as expressive language, executive function, and autism characteristics, we can gain insight into their interrelatedness and advance the understanding of the distinct, yet complex social behavioral phenotype associated with DS.

The purpose of this study was to provide preliminary data on the SSiS to measure social skills in a large sample of children and adolescents with DS. To accomplish this, the aims were to: (1) describe social skills and related behaviors captured by the SSiS in youth with DS; and (2) examine associations between the SSiS and measures of related domains (i.e., autism characteristics, executive function, and expressive language as measured by vocabulary). We expected that social skills measured by the SSiS would be negatively associated with autism characteristics and executive function impairments and positively associated with expressive language. We expected that behavioral problems measured by the SSiS would be positively associated with autism characteristics and executive function impairments and negatively associated with expressive language.

2. Materials and methods

2.1. Participants

Participants were combined from two research studies—a study on language in DS at the University of Illinois Urbana-Champaign (UIUC) and a multi-site study on measuring cognitive constructs at Cincinnati Children's Hospital Medical Center (CCHMC) and Colorado State University (CSU). Both studies were approved by the respective institution's Institutional Review Board. Eligibility criteria for the UIUC study were that the child with DS was between 6 and 11 years old, spoke English as a native language, communicated primarily through speech, and was able to speak in at least 2- to 3-word phrases according to parent report. Eligibility criteria for the CCHMC/CSU study were that the child with DS was between 6 and 17 years old, had English spoken as the primary language at home, and had a parent-reported developmental age of at least 3 years in order to engage in neuropsychological testing. No children were excluded from participation at CCHMC/CSU based on parental report of developmental age.

To be included in the current analyses, participants were required to have complete data on the SSiS. Thirteen participants from CCHMC/CSU were excluded due to missing SSiS data. This resulted in a sample size of 124 (M age = 11.61, SD = 3.48), n = 40 from UIUC and n = 84 from CCHMC/CSU. See Table 1 for demographic characteristics of the sample.

TABLE 1 Participant demographic characteristics.

	Full sample n =124	UIUC site n =40	CCHMC/ CSU site n =84
Age in years	M = 11.61 (SD = 3.48)*	M = 8.55 (SD = 1.62)	M = 13.07 (SD = 3.17)
Sex			
Male	47.6% (n = 59)	35.0% (n = 14)	53.6% (n = 45)
Female	52.4% (n = 65)	65.0% (n = 26)	46.4% (n = 39)
Race and ethnicity			
White	83.9% (n = 104)	77.5% (n = 31)	86.9% (n = 73)
Black	8.1% (n = 10)	12.5% (n = 5)	6.0% (n = 5)
Asian	4.0% (n = 5)	0	6.0% (n = 5)
Other	3.2% (n = 4)	7.5% (n = 3)	1.2% (n = 1)
Unreported	0.8% (n = 1)	2.5% (n = 1)	0
Hispanic	5.6% (n = 7)	2.5% (n = 1)	7.1% (n = 6)
Non-Hispanic	93.5% (n = 116)	97.5% (n = 39)	91.7% (n = 77)
Unreported	0.8% (n = 1)	0	1.2% (n = 1)

*Independent samples t-test indicated statistically significant differences between sites ($p < 0.01$).

2.2. Study design

As part of the larger studies, caregivers completed a series of questionnaires, including the SSiS (Gresham and Elliott, 2008), SRS-2 (Constantino and Gruber, 2012), and the Behavior Rating Inventory of Executive Function, 2nd edition (BRIEF2; Gioia et al., 2015). The children with DS also completed a direct assessment battery for each study that included the Expressive Vocabulary Test, 2nd or 3rd edition (EVT-2; Williams, 2007; EVT-3; Williams, 2019) and an IQ test. All UIUC participants were administered the EVT-2 (n = 37 after excluding 3 participants due to examiner error); CCHMC/CSU participants were administered the EVT-2 (n = 12) or the EVT-3 (n = 72), depending on year of enrollment. As a descriptive measure of IQ, UIUC participants completed the Leiter International Performance Test, 3rd edition (Leiter-3; Roid and Miller, 2013) nonverbal IQ test, and CCHMC/CSU participants completed the abbreviated version of the Stanford-Binet Intelligence Scales, 5th edition (SB-5; Roid, 2003; See Table 2).

2.3. Measures

2.3.1. Social skills

The SSiS Parent Form (Gresham and Elliott, 2008) is a standardized, norm-referenced questionnaire that asks caregivers to rate their child's behaviors as they relate to everyday social interactions. The SSiS informs intervention by identifying a child's social skills strengths and difficulties in acquisition or performance, including the presence of challenging behaviors that affect social interaction. Caregivers rate the frequency of each social skill (46 items) or problem behavior (33 items) over the last 2 months as *Never*, *Seldom*, *Often*, or *Almost Always*. For the social skills items, caregivers also rate the perceived importance of the behavior for their child's development (*not important*, *important*, or *critical*), but these ratings are not factored into the composite scores.

The Social Skills subscales are *Communication* (pragmatic skills), *Cooperation* (helping others, sharing, and compliance), *Assertion* (requesting, initiating, and responding appropriately), *Responsibility* (showing respect of property and communicating with adults), *Empathy* (showing concern for others' feelings and perspectives), *Engagement* (joining and inviting others to join activities, making friends), and *Self-Control* (responding appropriately to conflict and compromising). The Problem Behaviors subscales are *Externalizing* (verbal or physical aggression), *Bullying* (hurting others physically or emotionally, excluding others), *Hyperactivity/Inattention* (fidgety, impulsive, and easily distracted), and *Internalizing* (anxious, sad, or lonely). There is also an

TABLE 2 Participant performance on study measures.

	Full sample *M (SD) range*	UIUC site *M (SD) range*	CCHMC/CSU site *M (SD) range*
SSiS Social Skills composite	91.94 (14.62) 49–123	94.25 (13.22) 58–123	90.85 (15.19) 49–123
SSiS Problem Behaviors composite	104.39 (12.06)* 82–136	110.28 (12.09) 84–136	101.58 (11.05) 82–136
SRS-2 total *T*-score[a]	59.96 (8.66) 42–86	61.38 (8.09) 44–83	59.26 (8.89) 42–86
SRS-2 Restricted Interests and Repetitive Behaviors *T*-score[a]	60.46 (11.15)* 43–96	64.70 (11.38) 44–96	58.37 (10.49) 43–90
SRS-2 Social Communication and Interaction *T*-score[a]	59.55 (8.33) 42–84	60.13 (7.71) 44–81	59.27 (8.66) 42–84
BRIEF2 Global Executive Composite[b]	59.65 (8.95)* 38–81	63.79 (8.00) 48–80	57.77 (8.77) 38–81
BRIEF2 Behavioral Regulation Index	58.76 (9.32)* 37–82	62.63 (8.33) 46–82	56.92 (9.25) 37–76
BRIEF2 Emotional Regulation Index	55.71 (10.08)* 39–82	60.43 (10.61) 40–82	53.46 (9.05) 39–79
BRIEF2 Cognitive Regulation Index[a]	59.85 (8.52)* 38–84	63.03 (7.48) 45–77	58.42 (8.61) 38–84
EVT-2/3 standard score[a,c]	62.91 (12.98) 20–94	64.68 (12.97) 41–94	62.13 (12.98) 20–89
Leiter-3 nonverbal IQ	–	59.13 (9.49) 36–75	–
SB-5 ABIQ	–	–	49.36 (5.61) 47–76

*Independent samples t-test indicated statistically significant differences between sites ($p < 0.01$).

[a] $n = 121$;

[b] $n = 122$;

[c] EVT-2: $n = 37$ UIUC and $n = 12$ CCHMC/CSU, EVT-3: $n = 72$ CCHMC/CSU.

SSiS = Social Skills Improvement System. SRS-2 = Social Responsiveness Scale, 2nd edition. BRIEF2 = Behavior Rating Inventory of Executive Function, 2nd edition. EVT-2/3 = Expressive Vocabulary Test, 2nd edition/3rd edition. Leiter-3 = Leiter International Performance Scale, 3rd edition. SB-5 ABIQ = Stanford-Binet Intelligence Scales, 5th edition, Abbreviated Battery Intelligence Quotient.

Autism Spectrum subscale (difficulty connecting with others, repetitive behaviors, and rigidity) composed of items that span across the Social Skills and Problem Behaviors domains.

Subscale scores were converted to categorical ‘Behavioral Levels’ based on the raw score distribution in the normative sample. ‘Below Average’ indicates scores that are more than 1 SD below the normative sample mean, ‘Average’ indicates scores within ± 1 SD from the mean, and ‘Above Average’ indicates scores that are more than 1 SD above the mean. Composite scores for the two domains, Social Skills and Problem Behaviors, were computed based on chronological age and sex. Composite scores have a mean of 100 and standard deviation of 15. For all score types, higher scores in the Social Skills domain indicate stronger skills, and higher scores in the Problem Behaviors domain indicate more impairment. The SSiS publishers report high internal consistency for the Parent Form (median reliability $\alpha = 0.94–0.96$ for composite scales; median reliability $\alpha = 0.83–0.87$ for subscales) for the age ranges represented in the current study. They also reported moderate to strong correlations with the Behavior Assessment System for Children, 2nd edition (BASC-2; Reynolds and Kamphaus, 2008) for the age ranges represented in the current study (SSiS Social Skills and BASC-2 Adaptive Skills $r = 0.62–0.66$; SSiS Problem Behaviors and BASC-2 Behavioral Symptoms Index $r = 0.80–0.82$). Reported test–retest reliability for the SSiS Parent Form is also strong (median $r = 0.87$ for composite scales; median $r = 0.83$ for subscales).

2.3.2. Autism characteristics

The SRS-2 School-Age Form (Constantino and Gruber, 2012) is a 65-item standardized caregiver-report autism symptom screener for children ages 2.5–18 years. Caregivers rate the presence and frequency of their child’s behaviors within the last 6 months. The SRS-2 has two domains that align with DSM-5 diagnostic criteria for autism spectrum disorder—Social Communication and Interaction (SCI; with subdomains of *Social Awareness*, *Social Cognition*, *Social Communication*, and *Social Motivation*) and Restricted, Repetitive Behaviors and Interests (RRB). Chronological age and sex normed *T*-scores ($M = 50$, $SD = 10$) were computed for the SCI and RRB; an overall T-score was also computed. Higher scores indicate more autism-like symptoms. The SRS-2 publishers report strong psychometric properties in the standardization sample of children with and without autism for the School-Age Form (internal consistency $\alpha = 0.95–97$). Strong psychometric properties have also been reported for the SRS-2 in a smaller sample of children and adolescents with DS (Schworer et al., 2021).

2.3.3. Executive functioning

The BRIEF2 School-Age (Gioia et al., 2015) is a 63-item standardized caregiver-report questionnaire of everyday executive functioning for children ages 5–18 years. Caregivers rate the frequency in which their child engages in different behaviors in three categories that span different domains. The Behavioral Regulation Index (BRI) includes the domains of *Inhibit* and *Self-Monitor*. The Emotional Regulation Index (ERI) includes the domains of *Shift* and *Emotional Control*. The Cognitive Regulation Index (CRI) includes the domains of *Initiate*, *Working Memory*, *Plan/Organize*, *Task-Monitor*, and *Organization of Materials*.

T-scores were computed for each domain score and for the BRI, ERI, and CRI based on chronological age and sex norms. A Global Executive Composite (GEC) was also computed from chronological age and sex. Domain, index, and composite T-scores are all on the same scale ($M = 50$, $SD = 10$); higher scores indicate more dysregulation of executive function. The BRIEF2 publishers report strong test–retest reliability ($r = 0.88$ for the GEC; $r = 0.82–0.89$ for the index scores) and high internal consistency reliability ($\alpha = 0.97$ for the GEC; $\alpha = 0.90–0.96$ for the index scores) for the Parent Form.

2.3.4. Expressive language: Vocabulary

The EVT-2 (Williams, 2007) and EVT-3 (Williams, 2019) are different editions of a standardized measure of expressive vocabulary normed for ages 2.5 – 90 + years. For both versions, examinees are shown a series of pictures and are asked to verbally label each picture. Specific item content and basal/ceiling rules were updated in the EVT-3. The version that participants were administered depended on the timing of their study entry. Age-normed standard scores ($M = 100$, $SD = 15$) were used in data analysis. The EVT-2 and EVT-3 publishers report strong test–retest reliability (0.95 and 0.88, respectively). The versions are also highly correlated ($r = 0.86$; Williams, 2019).

2.3.5. Cognition

The Leiter-3 (Roid and Miller, 2013) is a standardized test of nonverbal cognition normed for ages 3 – 75 + years. It is nonverbal in administration and in method of response; examiners use gestures and facial expressions to model instructions, and examinees use pointing and other gestures to indicate their response. All UIUC participants completed the four Leiter-3 subtests (*Figure Ground*, *Form Completion*, *Classification and Analogies*, and *Sequential Order*) that yield a nonverbal IQ composite score ($M = 100$, $SD = 15$). The Leiter-3 publishers report good internal consistency reliability across composite scores (0.94–0.98) and its validation against the nonverbal IQ portion of the Stanford-Binet, 5th edition ($r = 0.77$). The SB-5 (Roid, 2003) is a standardized test of cognition that includes both verbal and nonverbal ability and is normed for ages 2 – 85 + years. All CCHMC/CSU participants completed the abbreviated battery IQ (ABIQ).

2.4. Data analysis plan

To address Aim 1, we first examined the distribution of SSiS Social Skills and Problem Behaviors composite scores across the sample. Next, we examined the distribution of subscale raw scores by behavioral level (Below Average, Average, or Above Average). For Aim 2, we conducted correlational analyses between the SSiS and the SRS-2, BRIEF2, and EVT-2/3. For these analyses, we included the SSiS subscales to determine the extent to which the different subscales demonstrate shared vs. distinct characteristics. Missing data ($n = 3$ EVT-2 from UIUC; $n = 3$ SRS-2 from CCHMC/CSU; $n = 2$ BRIEF2 CRI and GEC from CCHMC/CSU) were excluded pairwise such that the cases were excluded only from the correlational analyses involving the missing variables.

3. Results

3.1. Aim 1: Describe social skills and related behaviors captured by the SSiS in youth with DS

Table 2 provides the SSiS Social Skills and Problem Behaviors composite scores for the sample; Figure 1 shows the distribution of these scores. For Social Skills, skewness was −0.44 and kurtosis was 0.66. For Problem Behaviors, skewness was 0.45 and kurtosis was −0.29.

Figure 2 provides the distribution of subscale raw scores across the behavioral level categories (i.e., Below Average, Average, or Above Average), with the normative sample shown as a reference group. In the SSiS normative sample, the behavioral level categories were built around the raw score distributions for each subscale such that 'Below/Above Average' indicates ≥ 1 SD from the mean. Thus, 16% of the normative sample had scores that fell in the Below/Above Average categories. In the current sample of children with DS, the Social Skills subscales for which *more than* 16% scored in the Below Average category (i.e., less developed skills) were Cooperation (19%, $n = 23$), Assertion (40%, $n = 50$), Responsibility (35%, $n = 43$), and Engagement (27%, $n = 33$). The Problem Behaviors subscales for which *more than* 16% of the sample scored in the Above Average category (i.e., more challenging behaviors or dysregulation) were Externalizing (19%, $n = 23$) and Hyperactivity/Inattention (32%, $n = 40$). Finally, a relatively large portion of children scored in the Above Average (i.e., more symptomatic) category for the Autism Spectrum subscale (31%, $n = 39$).

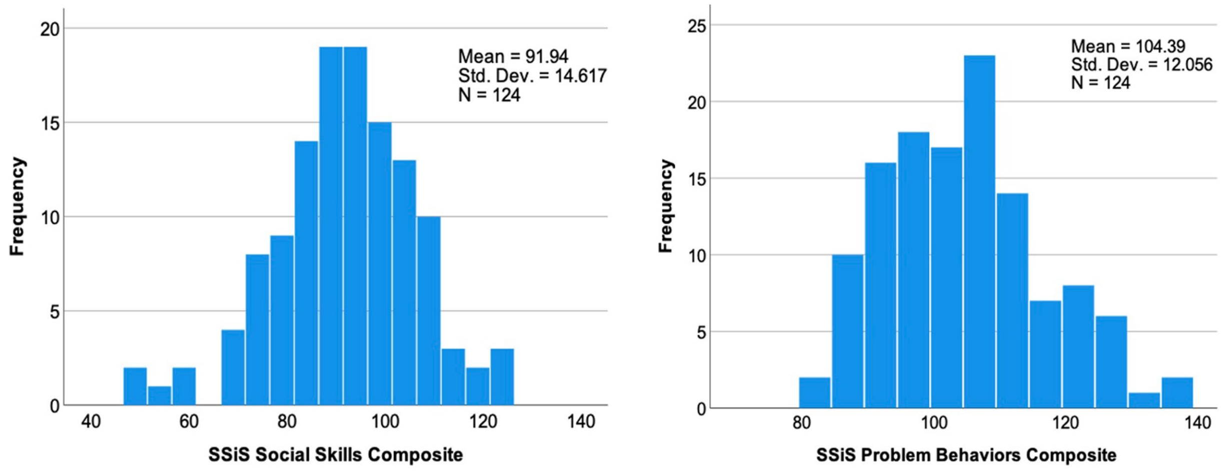

FIGURE 1
Distribution of Social Skills Improvement System (SSiS) composite scores.

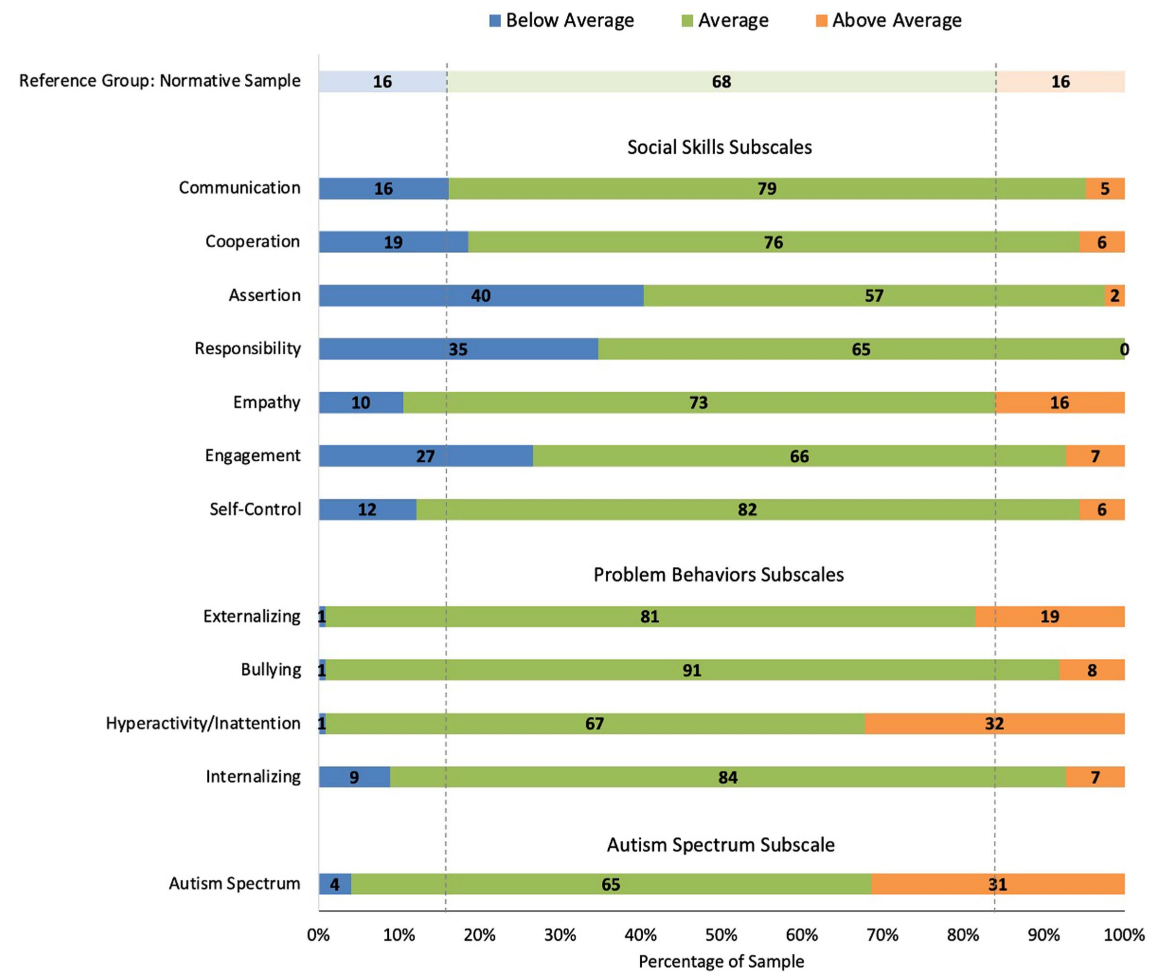

FIGURE 2
Percent of people with Down syndrome (DS) in each behavioral level category for the SSiS subscales.

3.2. Aim 2: Examine associations between the SSiS and measures of related domains

We conducted Pearson's *r* correlational analyses between SSiS composite standard scores, participant age, and composite/index/standard scores from the SRS-2, BRIEF2, and EVT-2/3. Correlations between these measures and SSiS Social Skills and Problem Behaviors are provided in Table 3. Overall, the SSiS Social Skills composite demonstrated large negative correlations with SRS-2 T-scores, moderate negative correlations with BRIEF2 Index scores, and a moderate positive correlation with EVT-2/3 standard scores. The SSiS Problem Behaviors composite showed large positive correlations with SRS-2 T-scores and BRIEF2 Index scores but was not significantly correlated with EVT-2/3 standard scores. Neither SSiS Social Skills nor Problem Behaviors composites were significantly correlated with participant age.

Correlations between the SSiS subscale raw scores and other measures are reported in Tables 4–6. Differential patterns emerged such that all the Social Skills subscales were significantly negatively correlated with SRS-2 scores and significantly positively correlated with EVT-2/3 scores; however, only some subscales were significantly correlated with different index scores of the BRIEF2 (Table 4), demonstrating differentiation across subdomains.

In contrast, all the Problem Behaviors subscales showed significant positive correlations with all BRIEF2 index scores, though strength of the correlations varied across subscales (Table 5). The Problem Behaviors subscales were also significantly positively correlated with SRS-2 scores, but no subscales were significantly correlated with EVT-2/3 scores. Finally, the Autism Spectrum subscale showed significant positive correlations across all SRS-2 and BRIEF2 scores as well as a significant negative correlation with EVT-2/3 scores (Table 6).

3.3. *Post hoc* analyses: Deeper characterization of the sample by SSiS behavioral level

Because a disproportionate number of children fell in the Below/Above Average behavioral level categories (i.e., compared to the normative sample) across several subscales, we conducted additional *post hoc* analyses to better characterize those participants. The goal was to subdivide the sample based on the SSiS behavioral levels and

TABLE 3 Pearson's *r* correlation coefficients for SSiS composite scores and other study measures.

	Age	SSiS SS	SSiS PB	SRS-2 total	SRS-2 RRB	SRS-2 SCI	BRIEF2 GEC	BRIEF2 BRI	BRIEF2 ERI	BRIEF2 CRI	EVT-2/3
Age	1	−0.040	−0.075	0.053	−0.024	0.069	−0.165	−0.178*	−0.088	−0.106	−0.177
SSiS Social Skills (SS)	–	1	−0.413**	−0.690**	−0.472**	−0.719**	−0.274**	−0.289**	−0.252**	−0.226*	0.359**
SSiS Problem Behaviors (PB)	–	–	1	0.631**	0.663**	0.572**	0.700**	0.604**	0.692**	0.574**	−0.107
SRS-2 total *T*-score	–	–	–	1	0.829**	0.977**	0.499**	0.452**	0.507**	0.408**	−0.218*
SRS-2 RRB *T*-score	–	–	–	–	1	0.696**	0.572**	0.536**	0.639**	0.424**	−0.253**
SRS-2 SCI *T*-score	–	–	–	–	–	1	0.435**	0.393**	0.421**	0.369**	−0.195*
BRIEF2 GEC	–	–	–	–	–	–	1	0.848**	0.789**	0.913**	−0.098
BRIEF2 BRI	–	–	–	–	–	–	–	1	0.635**	0.693**	−0.176
BRIEF2 ERI	–	–	–	–	–	–	–	–	1	0.580**	−0.138
BRIEF2 CRI	–	–	–	–	–	–	–	–	–	1	−0.034
EVT-2/3 standard score	–	–	–	–	–	–	–	–	–	–	1

*$p < 0.05$, **$p < 0.01$.

The shaded cells indicate significance at $p < 0.05$. SSiS = Social Skills Improvement System. SRS-2 = Social Responsiveness Scale, 2nd edition. RRB = Restricted, Repetitive Behaviors and Interests. SCI = Social Communication and Interaction. BRIEF2 = Behavior Rating Inventory of Executive Function, 2nd edition. GEC = Global Executive Composite. BRI = Behavior Regulation Index. ERI = Emotion Regulation Index. CRI = Cognitive Regulation Index. EVT-2/3 = Expressive Vocabulary Test, 2nd edition/3rd edition.

TABLE 4 Pearson's *r* correlation coefficients for SSiS Social Skills subscale raw scores and other study measures.

	Age	SSiS Comm	SSiS Coop	SSiS Assert	SSiS Resp	SSiS Emp	SSiS Engage	SSiS S-C	SRS-2 total T	SRS-2 RRB	SRS-2 SCI	BRIEF2 GEC	BRIEF2 BRI	BRIEF2 ERI	BRIEF2 CRI	EVT-2/3
Age	1	−0.134	0.133	−0.124	0.192*	0.009	−0.111	0.093	0.053	−0.024	0.069	−0.165	−0.178*	−0.088	−0.106	−0.177
SSiS Communication (Comm)		1	0.539**	0.636**	0.582**	0.659**	0.585**	0.518**	−0.566**	−0.328**	−0.611**	−0.187*	−0.183*	−0.133	−0.126	0.296**
SSiS Cooperation (Coop)			1	0.343**	0.831**	0.451**	0.346**	0.562**	−0.489**	−0.433**	−0.476**	−0.414**	−0.434**	−0.334**	−0.349**	0.258**
SSiS Assertion (Assert)				1	0.438**	0.483**	0.548**	0.419**	−0.537**	−0.279**	−0.589**	−0.018	−0.075	−0.036	0.012	0.288**
SSiS Responsibility (Resp)					1	0.525**	0.368**	0.583**	−0.528**	−0.425**	−0.531**	−0.359**	−0.408**	−0.296**	−0.272**	0.326**
SSiS Empathy (Emp)						1	0.571**	0.405**	−0.500**	−0.351**	−0.521**	−0.162	−0.168	−0.170	−0.079	0.248**
SSiS Engagement (Engage)							1	0.391**	−0.583**	−0.387**	−0.605**	−0.164	−0.094	−0.156	−0.187*	0.272**
SSiS Self-Control (S-C)								1	−0.443**	−0.331**	−0.456**	−0.297**	−0.307**	−0.380**	−0.182*	0.234**

*$p < 0.05$, **$p < 0.01$.

The shaded cells indicate significance at $p < 0.05$. SSiS = Social Skills Improvement System. SRS-2 = Social Responsiveness Scale, 2nd edition. RRB = Restricted, Repetitive Behaviors and Interests. SCI = Social Communication and Interaction. BRIEF2 = Behavior Rating Inventory of Executive Function, 2nd edition. GEC = Global Executive Composite. BRI = Behavior Regulation Index. ERI = Emotion Regulation Index. CRI = Cognitive Regulation Index. EVT-2/3 = Expressive Vocabulary Test, 2nd edition/3rd edition.

TABLE 5 Pearson's *r* correlation coefficients for SSiS Problem Behaviors subscale raw scores and other study measures.

	Age	SSiS Extern	SSiS Bully	SSiS Hyp/Inatt	SSiS Intern	SRS-2 total T	SRS-2 RRB	SRS-2 SCI	BRIEF2 GEC	BRIEF2 BRI	BRIEF2 ERI	BRIEF2 CRI	EVT-2/3
Age	1	−0.313**	−0.139	−0.321**	0.091	0.053	−0.024	0.069	−0.165	−0.178*	−0.088	−0.106	−0.177
SSiS Externalizing (Ext)		1	0.653**	0.855**	0.458**	0.425**	0.502**	0.369**	0.678**	0.642**	0.616**	0.517**	−0.011
SSiS Bullying (Bully)			1	0.463**	0.294**	0.262**	0.347**	0.219*	0.512**	0.460**	0.421**	0.353**	−0.083
SSiS Hyperactivity/ Inattention (Hyp/Inatt)				1	0.423**	0.507**	0.579**	0.448**	0.693**	0.646**	0.602**	0.556**	−0.019
SSiS Internalizing (Intern)					1	0.529**	0.420**	0.521**	0.398**	0.232**	0.463**	0.378**	0.015

$*p < 0.05$, $**p < 0.01$.

The shaded cells indicate significance at $p < 0.05$. SSiS = Social Skills Improvement System. SRS-2 = Social Responsiveness Scale, 2nd edition. RRB = Restricted, Repetitive Behaviors and Interests. SCI = Social Communication and Interaction. BRIEF2 = Behavior Rating Inventory of Executive Function, 2nd edition. GEC = Global Executive Composite. BRI = Behavior Regulation Index. ERI = Emotion Regulation Index. CRI = Cognitive Regulation Index. EVT-2/3 = Expressive Vocabulary Test, 2nd edition/3rd edition.

then examine patterns of characteristics across related domains (i.e., autism characteristics, executive function, and expressive language as measured by vocabulary). For these analyses, we focused only on the SSiS subscales for which more than 16% of the sample scored in the Below Average range for the Social Skills subscales or the Above Average range for the Problem Behaviors subscales. We converted composite/index/standard scores from the SRS-2, BRIEF2, and EVT-2/3 to *Z*-scores and plotted mean *Z*-scores for the SSiS Below/Above Average group relative to the rest of the participant sample.

Figure 3 shows performance across other study measures for the group of children with DS whose SSiS Social Skills subscale scores fell in the Below Average category relative to the remainder of the participant sample (i.e., those whose subscale scores fell in the Average or Above Average categories). For Assertion, the two subgroups diverged such that the 'Below Average' group had more elevated SRS-2 scores and lower EVT-2/3 scores. For Engagement, Cooperation, and Responsibility, the two subgroups diverged on all study measures. Overall, the participants in the 'Below Average' group for these three subscales had higher SSiS Problem Behaviors and higher (i.e., more symptomatic) SRS-2 and BRIEF2 scores. They also had lower EVT-2/3 scores. Finally, the participants in the 'Below Average' group for Engagement were older in age.

Figure 4 shows performance across other study measures for the group of children with DS whose SSiS Problem Behaviors and Autism Spectrum subscale scores fell in the Above Average category relative to the remainder of the participant sample (i.e., those whose subscale scores fell in the Average or Below Average categories). For Hyperactivity/Inattention, Externalizing, and Autism Spectrum subscales, the 'Above Average' groups showed lower SSiS Social Skills scores and higher (i.e., more symptomatic) SRS-2 and BRIEF2 scores. For Autism Spectrum, the participants in the 'Above Average' group also showed higher SSiS Problem Behaviors and lower EVT-2/3 scores. Finally, the participants in the 'Above Average' group for Hyperactivity/Inattention and Externalizing were also older in age.

4. Discussion

This study examined the SSiS in a large sample of youth with DS. The average social skills standard score across the sample was 92, falling well within 1 standard deviation from the mean of the normative sample. Similarly, the average problem behaviors standard score was 104. Upon examining behavior levels by subscale, most of the sample fell within the average range for each subscale. Thus, as a whole, youth with DS showed relatively mild symptoms associated with social interaction skills measured by the SSiS. This was surprising and conflicts with Guralnick et al.'s (2009, 2011) findings of significantly lower scores on the earlier version of this instrument, the SSRS, in 4- to 6-year-old children with DS. It also conflicts with the idea that as children with DS age and the demands of social interaction increase, they fall further behind their peers, especially in higher-order skills such as social cognition, social reasoning, and social problem-solving. However, the SSiS does not measure these individual skills but rather relies on informant report about behaviors that result from the coordination of many different social skills in real-world settings. Thus, it is possible that older children with DS develop compensatory strategies for navigating social interactions in naturalistic settings where more social cues are available. Regardless,

TABLE 6 Pearson's *r* correlation coefficients for SSiS autism spectrum subscale raw scores and other study measures.

	Age	SSiS Autism	SRS-2 total T	SRS-2 RRB	SRS-2 SCI	BRIEF2 GEC	BRIEF2 BRI	BRIEF2 ERI	BRIEF2 CRI	EVT-2/3
Age	1	0.002	0.053	−0.024	0.069	−0.165	−0.178*	−0.088	−0.106	−0.177
SSiS Autism Spectrum		1	0.791**	0.704**	0.764**	0.451**	0.365**	0.507**	0.314**	−0.290**

*$p < 0.05$, **$p < 0.01$.

The shaded cells indicate significance at $p < 0.05$. SSiS = Social Skills Improvement System. SRS-2 = Social Responsiveness Scale, 2nd edition. RRB = Restricted, Repetitive Behaviors and Interests. SCI = Social Communication and Interaction. BRIEF2 = Behavior Rating Inventory of Executive Function, 2nd edition. GEC = Global Executive Composite. BRI = Behavior Regulation Index. ERI = Emotion Regulation Index. CRI = Cognitive Regulation Index. EVT-2/3 = Expressive Vocabulary Test, 2nd edition/3rd edition.

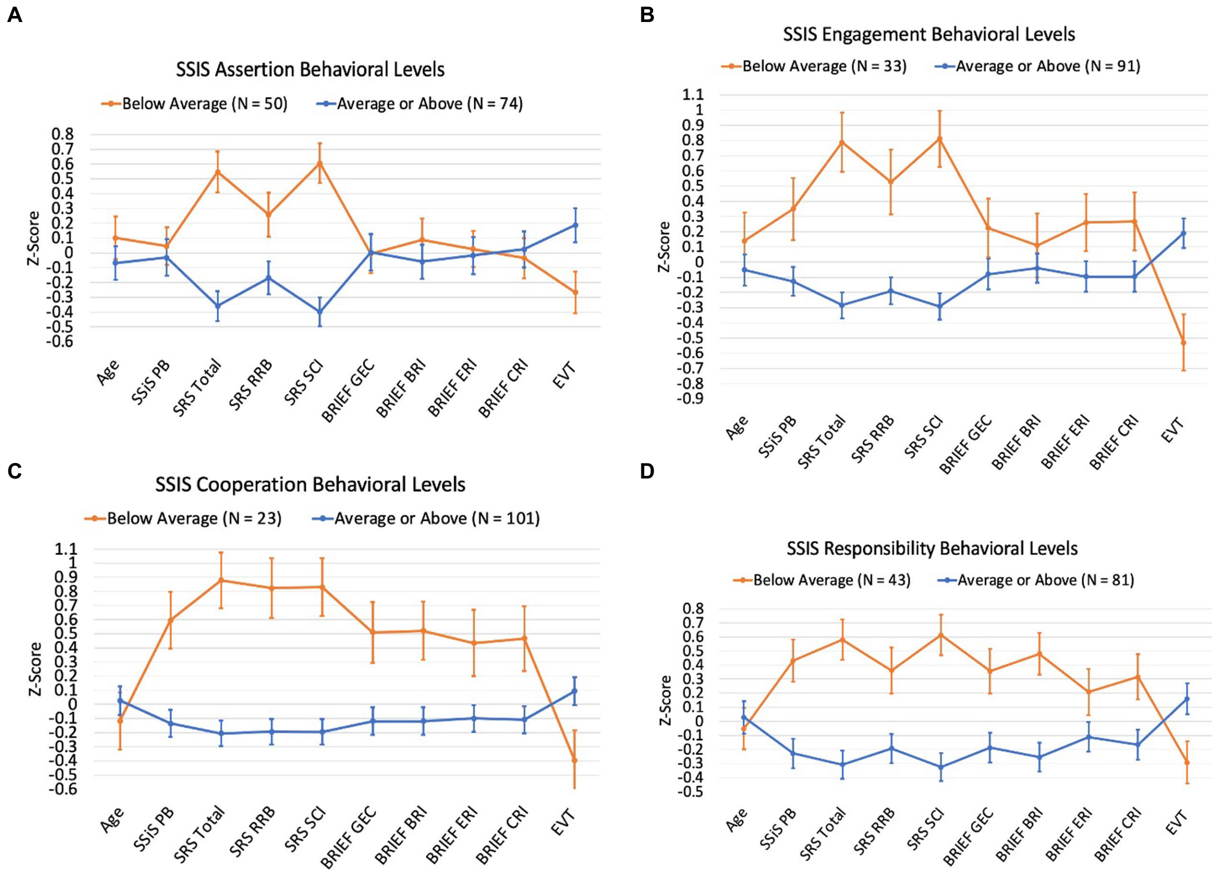

FIGURE 3
Participant scores across study measures by SSiS Social Skills subscale behavioral levels.

informant report measures such as the SSiS are important to consider clinically to gather meaningful information about social functioning across settings.

The current study provides some of the first evidence validating the use of the SSiS in youth with DS. SSiS composite scores correlated in the expected directions with SRS-2, BRIEF2, and EVT-2/3 composite scores of autism characteristics, executive function, and expressive vocabulary, respectively. Moreover, there was differentiation in the strength of these correlations such that EVT-2/3 expressive vocabulary was significantly and moderately correlated with SSiS social skills but only weakly correlated (failing to reach significance) with SSiS problem behaviors. Additionally, BRIEF2 executive function impairments were strongly correlated with SSiS problem behaviors and moderately correlated with SSiS social skills. Further, differentiation in the strength of associations between the SSiS subscales and subdomain/index scores on the SRS-2 and BRIEF2 provide initial evidence of construct validity, although this should be systematically tested through future research.

Across the sample, participants showed different distributions of low/average/high scores by subscale. That is, for some subscales, a disproportionate amount of the sample scored in the below average (i.e., lower levels of social skills) or above average (i.e., more symptomatic in problem behaviors or autism spectrum) range relative to the normative sample; for other subscales, the sample's score distribution aligned more closely to that of the normative sample. For social skills, the subdomain with the greatest proportion of the sample scoring in the below average range was *Assertion*. These items refer to skills such as initiating a conversation, asking for help, and speaking up for oneself. The next subscale with a disproportionate amount of the sample in the below average range was *Responsibility*, which

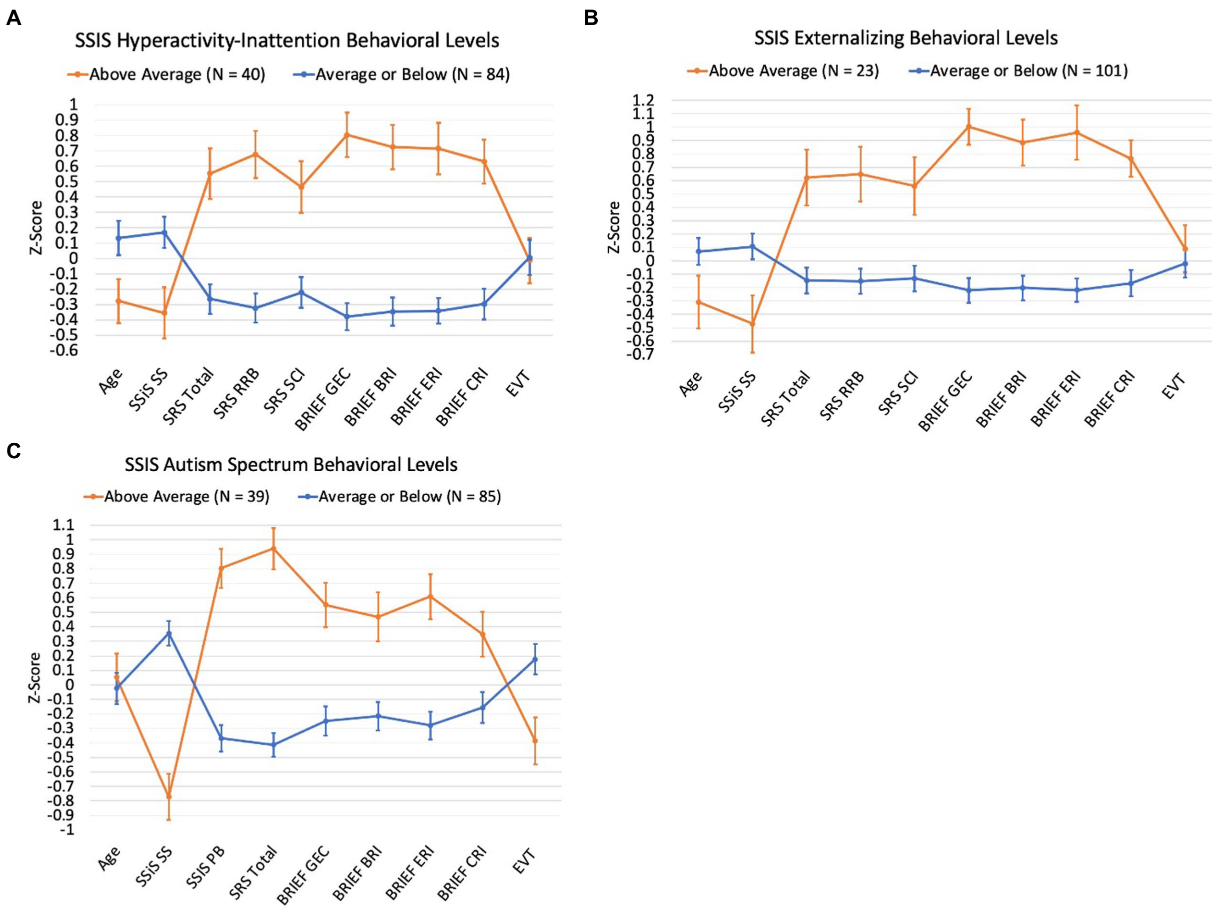

FIGURE 4
Participant scores across study measures by SSiS Problem Behaviors and Autism Spectrum subscale behavioral levels

includes items related to taking responsibility for one's own actions, following through, and showing respect for others' property. The next subscale in the below average range was *Engagement*, which includes initiating and joining interactions with peers, making friends, and starting conversations with others. The final social skills subscale that showed only slightly more children in the below average range relative to the normative sample was *Cooperation*, which mostly asks about the home environment—following rules or caregiver instructions and getting along at home. In contrast, the subscales for which the sample with DS more closely aligned with the normative sample were *Empathy*, which includes showing concern for others and trying to understand their feelings, *Communication*, which includes mostly nonverbal pragmatic skills like turn-taking, appropriate tone and eye contact, and gesturing, as well as *Self-Control*, which includes staying calm when others are aggressive or disagree and compromising.

Aspects of this profile fit broadly with what is known about the DS phenotype. For example, during experimental or language sampling tasks, children with DS have initiated conversational topics and signaled their own non- comprehension to repair conversational breakdowns less often than developmental age-matched typically developing peers (see Martin et al., 2009 for a review; Martin et al., 2018). This aligns with the lower scores observed for *Assertion* and *Engagement* in the current study. The findings for *Empathy* and *Communication* in the current sample align with observations of relatively strong empathic and pro-social behaviors noted by Kasari et al. (2003) and relative strengths in nonverbal communication noted in studies of early social development in young children with DS (see Cebula et al., 2010 for a review). The current study extends prior research by characterizing a profile of relatively more and less impaired social skills used during everyday social interaction by children and adolescents with DS.

For problem behaviors, the subdomain with the highest proportion of the sample scoring in the above average range was *Hyperactivity/Inattention*. These items refer to impulsive behaviors and interrupting, fidgeting, distractibility, and temper tantrums. Slightly more children scored in the above average range for *Externalizing*; interestingly, many of the *Hyperactivity/Inattention* subscale items feed into this subscale, but it also includes unique items related to disobedience and defiance. The subscale scores for which the sample with DS more closely aligned with the normative sample were *Bullying* and *Internalizing*. These findings correspond with other research pointing to particularly high rates of hyperactivity, inattention, and noncompliance among youth with DS (Capone et al., 2006; Jacola et al., 2014; Patel et al., 2018; Esbensen et al., 2021) along with slightly elevated aggressive behaviors (van Gameren-Oosterom et al., 2011) and relatively low rates of internalizing symptoms, at least during childhood (van Gameren-Oosterom et al., 2011, 2013; Channell et al., 2019). Interestingly, both *Hyperactivity/Inattention* and

Externalizing scores were negatively correlated with age, indicating a decrease in symptoms across age in the current sample. Broadly speaking, this fits with others' reports of age-related differences in the pattern of maladaptive behaviors in DS across childhood and adolescence (Dykens, 2007; van Gameren-Oosterom et al., 2011, 2013).

Finally, 31% of the sample scored in the above average range for the *Autism Spectrum* subscale, which includes both social skills and problem behaviors commonly associated with features of autism. This is largely consistent with other findings that children with DS tend to show elevated scores on autism screeners and symptom monitoring measures that capture broad autism-like characteristics (DiGuiseppi et al., 2010; Warner et al., 2014; Channell et al., 2015; Channell, 2020).

It is also important to recognize the variability observed across this study's sample in SSiS composite standard scores and in the distribution of scores across subscales reported in Figures 1 and 2. That is, a range of scores were observed for the SSiS social skills and problem behaviors composites and in the distribution of scores across subscales. Additionally, we plotted participants' sores on the other study measures of autism characteristics, executive function, and expressive vocabulary, with separate plots for the subsample of participants who scored in the below/above average (social skills/ problem behaviors) on a subscale relative to the remainder of the sample (see Figures 3, 4). These data reveal systematic differences among subsamples of participants who show greater impairments vs. those who show average or better social skills and problem behaviors. These results support Channell et al.'s (2021) findings of potential subgroups within the DS phenotype. However, the current study did not use latent profile analysis, and the extent to which the same participants who fell in the below/above average category across the different subscales is unknown. Regardless, these data demonstrate the importance of considering individual differences within the DS phenotype (Karmiloff-Smith et al., 2016).

4.1. Limitations and future directions

Although the current sample size was large, the study did not employ an epidemiological design. Therefore, one should not interpret the SSiS mean composite scores or percentages of children falling into the behavioral levels as such. More work is needed to determine the extent to which these findings generalize (or do not generalize) to the larger population with DS. Based on other measures included in this study, the current sample appears largely similar to what is reported in the literature about DS in terms of IQ (see Grieco et al., 2015), executive function (e.g., Loveall et al., 2017; Rosser et al., 2018), and autism characteristics (Channell et al., 2015); however, convenience sampling is a common issue in DS research. Additionally, the current sample was 84% White and 94% Non-Hispanic, much less diverse than recent United States population-based data on DS reported by Mai et al. (2019; 62% Non-Hispanic and 43% White Non-Hispanic) and United States Census estimates (76% White and 81% Non-Hispanic; U.S. Census Bureau, 2022), although race and ethnicity were categorized differently in these reports than in the current study. Thus, it will be important to examine the utility of the SSiS across more culturally diverse samples in which cultural expectations for social interaction may also differ. Furthermore, we combined data from two different studies to yield a larger sample size; however, both studies used different inclusion/exclusion criteria. Notably, the UIUC study required that participants were able to speak in phrases and use spoken language as their primary mode of communication, who may also be more advanced developmentally. Thus, it is possible that we had an overrepresentation of individuals with DS with more advanced skills in the current sample. Further, both studies required that participants could complete an in-person assessment battery. This criterion likely led to the exclusion of children with DS with more maladaptive behaviors and/or lower developmental levels from these studies. Such children may have more limited social interaction skills or a different social skill profile than what was observed in the current sample, again limiting generalizability to the broader population with DS. Future research should focus on examining the SSiS in a more inclusive sample of children with DS, both developmentally and demographically.

4.2. Conclusion

Very few measures of social skills have been established for youth with DS, particularly for older youth who experience increased social demands. The SSiS is a social skills assessment tool that is useful clinically because it provides information about how an individual functions during day-to-day social interactions across different settings. The results provide preliminary data on the SSiS in youth with DS and contribute some of the first evidence validating its use in this population. This study also informs the understanding of the DS phenotype by using the SSiS to report social skills profiles across a sample of children and adolescents with DS in relation to autism characteristics, executive function, and expressive language. Moving forward, a well-researched tool for measuring everyday social interaction skills in this population will advance the understanding of this aspect of the DS phenotype and will provide clinicians with an assessment tool for diagnosing, intervention planning, and treatment monitoring.

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 Illinois Urbana-Champaign, Cincinnati Children's Hospital Medical Center, and Colorado State University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

MC, LM, and AE conceptualized this study. MC, DF, and AE led the original studies from which data were used in this study. MC led manuscript drafting and conducted data analysis. MC, LM, ES, DF, and AE edited the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Joint engagement and early language abilities in young children with Down syndrome

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Introduction: Early social strengths likely serve as a foundation for language acquisition for young children with Down syndrome (DS). One way to characterize early social skills is to examine a child's engagement with a caregiver around an object of interest. The current study examines joint engagement in young children with DS and its relation to language abilities at two-time points in early development.

Methods: Participants were 16 young children with DS and their mothers. At two time points, mother–child free plays were completed and coded for joint engagement. Language abilities were measured at both time points using the Vineland Adaptive Behavior Scales 3rd edition and the number of words understood and produced on the MacArthur-Bates Communication Development Inventory.

Results: Young children with DS spent more time in supported joint engagement than coordinated joint engagement at both time points. Using a weighted joint engagement variable, children with DS who had higher weighted joint engagement had lower expressive language raw scores on the Vineland when controlling for age at Time 1. At Time 2, children with DS who had higher weighted joint engagement had higher expressive and receptive language raw scores on the Vineland when controlling for age. Predictively, children with DS who had a higher weighted joint engagement at Time 1 had a lower number of words produced at Time 2 when controlling for age at Time 1.

Discussion: Our results suggest that young children with DS may compensate for their difficulties with language by using joint engagement. These results highlight the importance of teaching parents to be responsive during interactions with their child to move them into both supported and coordinated engagement, which in turn may foster language development.

KEYWORDS

Down syndrome, joint engagement, language development, caregiver-child interaction, joint attention

1. Introduction

Early social skills likely serve as a foundation for language acquisition for young children with Down syndrome (DS). While early social strengths are a hallmark feature of DS [e.g., social orienting, directing eye gaze, vocalizations, gestures, social engagement, and empathy; (Fidler, 2005, 2006; Fidler et al., 2008; Hahn, 2016)], there has been little research examining the relationship between early social skills and language development in this population. One way to characterize early social skills is to examine a child's engagement with people, objects, and events during a naturalistic interaction (Bakeman and Adamson, 1984; Adamson and Bakeman, 1991;

Adamson and Chance, 1998; Adamson et al., 2004). Joint engagement is used to describe periods or episodes of joint attention (Bakeman and Adamson, 1984; Adamson and Chance, 1998; Adamson et al., 2004). That is, joint engagement usually describes the quality of an interaction for a duration of time (Adamson and Bakeman, 1991). In contrast, joint attention is often described, especially as measured in the extant literature, as a point estimate or count that is then used to calculate a rate of joint attention. Joint engagement adds more nuance to joint attention because it differentiates the emergence of joint attention, referred to as supported joint engagement, and the consolidation of skills needed to participate in joint attention interactions with another person, referred to as coordinated joint engagement (Hahn, 2016). Because joint engagement is the foundation for later language development (Smith et al., 1988; Bopp and Mirenda, 2011) understanding this relationship has implications for early language intervention targets and timing. Thus, the purpose of the present study was to examine joint engagement and its relation to language abilities in young children with DS across two-time points.

Joint engagement emerges when a child begins to include a caregiver in their interaction with an object. Before this, children's engagement, or active attention, is more solely focused on either a person or an object (Adamson and Bakeman, 1991). But as their attention and shifting abilities increase, they begin to share their interests with another person. Because children are learning to consolidate the skills needed to actively share attention (i.e., using eye gaze, affect, gestures, and vocalizations to indicate their attention and interest; Adamson and Bakeman, 1991; Adamson and Chance, 1998), joint engagement is often divided into two forms: supported joint engagement and coordinated joint engagement. Instances of supported joint engagement occur when the child and the caregiver are engaged with the same object, but the caregiver is scaffolding the interaction (i.e., gesturing toward the object or talking about the object) while the child may not consistently reciprocally respond to the caregiver because they are still learning to share their attention (Bakeman and Adamson, 1984; Adamson and Bakeman, 1991; Adamson et al., 2004). Thus, supported joint engagement represents the emergence of the ability to share attention (i.e., joint attention) during which the child needs support from their caregiver to sustain the interaction. This support leads to the ability to share their attention actively and reciprocally with a caregiver (i.e., to use joint attention effectively). Therefore, coordinated joint engagement occurs when the child has gained the skills to share attention and actively engage with the object and caregiver in a dynamic and reciprocal interaction where they are initiating and responding to the actions of the other person around the object (Bakeman and Adamson, 1984; Adamson et al., 2004). It is important to note that although there is a developmental progression of these different engagement states—person, object, supported, coordinated—children will continue to use early forms of engagement even once skills are consolidated based on the demands of the social situation (see Table 1 for definitions and examples of each type of engagement; Bakeman and Adamson, 1984; Adamson and Chance, 1998; Hahn et al., 2016).

Engagement states in neurotypical children emerge over the first 2 years of life. Infants begin to engage with their environment within the first month (Adamson and Chance, 1998; Trevarthen and Aitken, 2001). By the second month, infants begin to participate in face-to-face interactions with their caregiver, which is the emergence of person engagement (Adamson and Chance, 1998; Trevarthen and Aitken, 2001). Periods of person engagement start to decrease between 5 and 6 months as infants begin to focus their attention on objects in their environment (i.e., object engagement; Trevarthen and Hubley, 1978; Adamson and Chance, 1998). At this time infants are not yet able to coordinate their interest in objects with the caregiver. However, as infants learn to consolidate their attention and shifting skills, caregivers join in the infant's engagement with an object leading to the emergence of supported joint engagement after 6 months (Bakeman and Adamson, 1984; Adamson and Bakeman, 1991; Adamson and Chance, 1998). Initially, most interactions between the infant, caregiver, and an object of interest are prolonged periods of supported joint engagement, but as the infant continues to consolidate their attention and shifting skills moments of coordinated joint engagement begin to emerge (Bakeman and Adamson, 1984; Adamson and Chance, 1998). By 12–13 months infants can engage in sustained periods of coordinated joint engagement and by the middle of the second-year infants can use coordinated joint engagement with ease (Bakeman and Adamson, 1984; Adamson and Chance, 1998).

Although most of the research on joint engagement has been conducted in neurotypical children, there is a small body of research in DS. Broadly, research on joint attention and joint engagement in DS often discusses this as an area of relative strength; however, a recent meta-analysis that included studies of both joint attention and joint engagement indicated that these skills, while not a weakness, appear to be commensurate with developmental level (Hahn et al., 2018). Further, the results of this meta-analysis suggest that joint attention and joint engagement may be a strength relative to other aspects of the DS behavioral phenotype and when compared to those with other neurodevelopmental disabilities. Nonetheless, given the small number of studies on joint engagement in DS, it is difficult to draw conclusions about the pattern of performance across engagement statements and the emergence of these skills. For example, Legerstee and Weintraub (1997) found that compared to mental-age-matched neurotypical peers, 8-to-32-month-olds with DS spent more time in supported joint engagement and less time in object engagement and coordinated joint engagement on average across four-time points. In contrast, another study noted that 30-month-olds with DS spent similar amounts of time in supported joint engagement as 18-month-old neurotypical peers (Adamson et al., 2009). Further, in this study, children with DS also spent less time in object engagement and slightly more time in coordinated joint engagement than neurotypical 18-month-olds (Adamson et al., 2009). However, in another study, 20-to-68-month-olds with DS spent more time in coordinated joint engagement and a similar amount of time in supported joint engagement as compared to neurotypical peers matched on receptive vocabulary (Lewy and Dawson, 1992). In a study of Italian 24-month-olds with DS using a similar, albeit not the same coding scheme as other studies of joint engagement, children spent more time in joint attention (i.e., coordinated joint engagement) and less time in passive attention (i.e., supported joint engagement; Zampini et al., 2015). The variability across these studies demonstrates that the use of these skills is fluid and evolving. That is a child can continue to use a less complex engagement state depending on the demands of the situation and they are in the process of learning to use more complex states more competently and efficiently leading to variability in performance. Importantly, there is evidence that coordinated joint engagement increases with development in DS (Legerstee and Weintraub, 1997; Adamson et al., 2009), but at a slower rate than mental-age matched neurotypical peers (Legerstee and Weintraub, 1997). Again, this highlights that the development of joint engagement is progressing.

Examination of language outcomes related to joint engagement in DS has only focused on the most complex forms of supported and coordinated joint engagement in which the child *also* needs to include spoken communication to comment during the interaction (referred to as symbol-infused supported and coordinated joint engagement; Adamson et al., 2004, 2009). While this suggests that the amount of time in symbol-infused supported and coordinated joint engagement accounts for variance in both expressive and receptive vocabulary scores in 30-month-olds with DS (Adamson et al., 2009), it is still unclear how supported and coordinated joint engagement where the child is *not yet* incorporating spoken communication or symbols supports language abilities in DS. Given the variability in the onset of first words in DS (9–24 months; Martin et al., 2009), moments of supported and coordinated joint engagement may be ideal moments for language learning as evidenced by research on joint engagement and joint attention and language in DS (Zampini et al., 2015; Seager et al., 2018) and the use of joint engagement in language interventions for children with autism (Kasari et al., 2008, 2010, 2012). Thus, the present study seeks to describe engagement states in young children with DS across two-time points. In addition, we examined the relationship between joint engagement and language abilities concurrently and predictively. Our research questions were:

1. What is the pattern of engagement for young children with DS at Time 1 and Time 2?
   a. Do young children with DS spend different amounts of time in supported joint engagement than coordinated joint engagement at Time 1 and Time 2?
2. What is the relationship between joint engagement and expressive and receptive language ability concurrently and predictively (i.e., joint engagement at Time 1 to language measures at Time 2)?

2. Materials and methods

2.1. Participants

Participants were 16 young children with DS (9 males, 7 females) and their mothers (*M* age = 42 years, SD = 5.29, range 20–42 years). At Time 1, children were between 12–30 months, and at Time 2, they were between 21–38 months (see Table 2 for child characteristics). Most children were White (62.5%), 31.3% were more than one race, and 6.3% were Asian. For family income, 31.5% were between $20,000–$50,000, 31.4% were between $50,001–$100,000, 31.3% had incomes of $100,001 or above, and 6.3% choose not to report their family income. Most mothers had attended some college (43.8%), 25% graduated college, and 31.3% had a graduate or professional degree.

Participants were drawn from two studies examining early language development in DS (12–24 and 18–30 months, respectively) conducted by the first author (see Table 2 for information about language use). For both studies, participants were recruited from the Midwest and Southern regions of the United States through flyers shared with local DS parent groups and early intervention service providers either through email, social media, or newsletters. All children with DS were reported to have normal or corrected hearing and vision, and English was the primary language spoken in their homes.

2.2. Procedures

All study procedures were approved by the Institutional Review Board at the University of Illinois at Urbana-Champaign. As part of a larger assessment battery for each study, children and their mothers completed a 15-min free play with a set of developmentally appropriate toys and were instructed to play as they normally would. For the study of children between 12 to 24 months, toys included stacking rings, a set of sensory balls, board books, a cloth book, rattles, a shape sorter, stacking blocks, and connecting rings. For the study of children 18 to

TABLE 1 Definitions and examples of engagement states.

Engagement State	Definition	Example
Unengaged	The child is not interacting with objects or the caregiver.	The child is scanning the room.
Person	The child is exclusively interacting with the caregiver in a face-to-face interaction without involving objects or toys.	The child's body is positioned toward the caregiver and involved in face-to-face interactions (e.g., peek-a-boo, patty cake, etc.).
Object	The child is playing with an object alone, interacting only with the object, and not interacting with or including the caregiver in their play.	The child is focused on building a tower by staking a set of blocks.
Supported Joint Engagement	The child and the caregiver are interacting with the same object, but the child is not actively acknowledging or responding to the caregiver's participation.	The child and the caregiver are playing with a shape sorter, and the mother encourages the child by commenting or asking questions about the object such as "Green square," or "Where does that piece go?" or supports the child's play by handing the child the shapes to insert in the slots. The child might briefly acknowledge or respond to the caregiver but not consistently or for an extended period.
Coordinated Joint Engagement	The child and the caregiver are interacting with the same object, and the child repeatedly acknowledges and responds to the caregiver's participation in the interaction between the two of them and the object.	The child and the caregiver are playing with a puzzle and the child points to a puzzle piece of a dog and then looks at the caregiver. The caregiver says, "can you put the dog in?." The child tries to put the piece in place using eye gaze and facial affect to check in with the caregiver.

TABLE 2 Participant characteristics.

Characteristic	Time 1			Time 2		
	M	SD	Range	*M*	SD	Range
Child						
Age in months	19.63	4.65	12–30	28.31	5.07	22–38
Vineland Adaptive Behavior Composite	69.07	10.73	45–81	69.81	10.37	51–89
Vineland receptive language raw score	22.75	11.66	4–52	30.19	10.77	11–53
Vineland expressive language raw score	15.81	7.07	4–32	18.88	8.36	5–32
Number of words understood (CDI)	96.87	95.64	6–327	159.87	107.06	28–318
Number of words produced (CDI)	6.27	7.16	0–25	22.47	23.47	1–93

30 months, toys included stacking boxes, animal figurines, a ball, a rattle, connecting rings, board books, a cloth book, an animal puzzle, a teddy bear, 2 plastic bowls, plastic spoons, and plastic forks. Mothers were administered the Vineland Adaptive Behavior Scales 3rd edition (VABS; Sparrow et al., 2016) and completed the MacArthur-Bates Communicative Development Inventory – Words & Gestures (CDI; Fenson et al., 2006). The COVID-19 pandemic interrupted the study for 18-to-30-month-olds and required a transition from in-person assessment to remote assessment. Before the COVID-19 pandemic, participants were visited in person in their homes (Time 1 n = 11; Time 2 n = 9). Families who either enrolled in this study after March 2020 or were due to be seen for their second timepoint after March 2020 (Time 1 n = 5; Time 2 n = 7), completed the free play and were interviewed with the VABS over Zoom. Independent samples *t*-tests indicated no significant differences between those who participated in-person as compared to those who participated remotely on supported or coordinated joint engagement at either time point. Data were managed in REDCap electronic data capture tools hosted at the University of Illinois (Harris et al., 2009, 2019).

2.3. Measures

2.3.1. Joint engagement

Joint engagement was coded based on a coding scheme developed by the first author (Hahn et al., 2016; Mattie and Hadley, 2021) based on the coding scheme developed by Bakeman and Adamson (1984). Joint engagement was coded from video recordings of the 15-min free play at both time points. Mothers were asked to play with their child as they normally would. Table 1 provides the definitions and an example of each engagement state. Codes were based on the *child's* engagement during the interaction. Therefore, the coder was identifying what/whom the child was engaged with to differentiate supported and coordinated joint engagement based on how they were engaging. A full description of the coding scheme is available in Mattie and Hadley, 2021. See Table 1 for definitions and examples of each engagement state.

Video recordings were digitized and coded using Noldus The Observer XT 14 software (Noldus The Observer XT, 2016). Coders would watch the videos in real-time coding for when an engagement state would start and end. An engagement state was defined as "a period of at least 3 s that is characterized by the child's active interest in people and in objects and events" (Adamson et al., 2004, p. 1,176). To identify the start and end of an engagement state, coders would look for a breakpoint (Newtson, 1973; Bakeman and Adamson, 1984) in the interaction between the child and the mother. When a breakpoint was noted, coders would rewatch the video until they felt they had accurately identified the breakpoint. They would also check that the engagement state lasted for at least 3 s to ensure that it met the criteria for an engagement state.

2.3.1.1. Coder training and reliability

The first author trained the second author on the coding scheme by explaining the scheme in-depth and providing examples of the behavior to be coded by watching videos together. The second author then served as the primary coder for all videos. Reliability was conducted on 8 randomly assigned videos (25% of the video data), which were coded by the first author. Intraclass correlation coefficients (ICCs; Shrout and Fleiss, 1979) were calculated between the primary and reliability coder for the length of time in each engagement state. For each state, the ICCs were unengaged 0.90, object 0.57, SJE 0.76, and CJE 0.85. The ICC for object engagement is lower than the others, this is due to difficulty identifying if there were 3-s of time of object engagement and separating when the state moved from object engagement into either supported or coordinated joint engagement. That is, children were often interested in interacting with their mother, which resulted in either supported joint engagement, due to the mother joining and scaffolding their engagement with the toy or coordinated joint engagement because the child initiated and maintained an active interaction with their mother and the toy. Person engagement rarely occurred in the present study (see Table 3), with only 7 participants using this state; therefore, an ICC was not calculated.

2.3.2. Language abilities

Language abilities were measured at both time points using a functional measure (Vineland Adaptive Behavior Scales 3rd edition, VABS) and a parent-reported count of words understood and words produced (MacArthur-Bates Communication Development Inventory, CDI).

2.3.2.1. Vineland Adaptive Behavior Scales 3rd edition, comprehensive interview

The VABS is a standardized caregiver interview of adaptive functioning across three domains: communication, socialization, and daily living (Sparrow et al., 2016). The present study examined the receptive and expressive communication subdomains at each time point to measure functional communication. Items on the VABS are scored by the interviewer on a 3-point Likert scale describing the individual's ability to do different functional skills independently (*not yet, sometimes, usually/always*). The VABS has well-established reliability and validity.

TABLE 3 Frequency and mean proportion of time spent in each engagement state.

Engagement state	Time 1				Time 2			
	Number of children who engaged in each state	Mean proportion of time	SD	Range	Number of children who engaged in each state	Mean proportion of time	SD	Range
Unengaged	15	0.09	0.11	0.00–0.38	15	0.07	0.06	0.00–0.20
Person	7	0.01	0.02	0.00–0.05	7	0.02	0.05	0.00–0.15
Object	16	0.26	0.12	0.10–0.46	16	0.28	0.13	0.09–0.50
Supported Joint Engagement	16	0.52	0.17	0.19–0.77	16	0.53	0.16	0.21–0.76
Coordinated Joint Engagement	11	0.07	0.06	0.00–0.20	9	0.05	0.05	0.00–0.16
Weighted Joint Engagement	16	0.69	0.24	0.19–1.08	16	0.62	0.19	0.21–0.97

2.3.2.2. MacArthur-Bates Communicative Development Inventories-Words and Gestures

The CDI is a standardized caregiver report of early communication that provides a checklist of 396 common words their child understands and/or produces *via* speech (Fenson et al., 2006). Although the CDI is standardized, raw scores are not transformed into standard scores. Therefore, if a caregiver reports their child understands 50 words, this number represents the final score. For the present study, language abilities were measured by the number of words understood (receptive vocabulary) and the number of words produced (expressive vocabulary). The CDI has well-established reliability and validity.

2.3.3. Developmental level

Developmental level was measured using the overall level of adaptive functioning. Adaptive functioning was measured by using the Adaptive Behavior Composite from the VABS ($M = 100$, $SD = 15$) (Sparrow et al., 2016).

2.4. Data reduction

The data extracted from Noldus the ObserverXT was used to calculate the proportion of time spent in each engagement state in seconds by taking the mean duration each participant spent in each state and dividing it by the total time of the observation. This approach was used because although the average length of the mother–child free play was 15 min (i.e., there were slight variations in the total time of each participant's observation Time 1: $M = 913.78$ s, $SD = 86.88$ s; Time $2 = M = 928.98$ s; $SD = 29.86$ s).

We also calculated a weighted joint engagement score to indicate each child's level of joint engagement development. This approach accounts for growth and the increasing complexity of joint engagement behaviors (see Hahn et al., 2016). Similar approaches have been used to examine increases in early communication (Luze et al., 2001; Greenwood et al., 2003) and differentiate levels of play complexity (Thiemann-Bourque et al., 2012). Weighted joint engagement was calculated by rank ordering behavior from less to more complex (i.e., supported joint engagement = 1, coordinated joint engagement = 2); therefore, we multiplied each child's coordinated joint engagement score by 2 and added the supported joint engagement score of each child (Hahn et al., 2016). For example, if the proportion of time spent in coordinated joint engagement was 0.20 and the proportion of time spent in supported joint engagement was 0.30, then the weighted joint engagement score would be 0.70 (i.e., $[0.20 \times 2] + 0.30 = 0.70$; see Table 3 for means, standard deviations, and ranges).

2.5. Data analysis

For our first research question, descriptive statistics were used to explore the pattern of engagement at Time 1 and Time 2. Next, we used paired-sample *t*-tests to examine if there were differences between supported and coordinated joint engagement at each Time 1 and Time 2. The proportion of time spent in each engagement state was used for these analyses.

For our second research question, we used partial correlations controlling for child chronological age to examine the relationship between joint engagement and language abilities concurrently and predictively (receptive and expressive raw scores from the Vineland and the number of words understood and the number of words produced from the CDI). For these analyses, the weighted joint engagement score was used. For the predictive correlations weighted joint engagement at Time 1, controlling for chronological age at Time 1, and language abilities at Time 2 were used.

3. Results

3.1. Pattern of engagement

Young children with DS, on average, spent the most time in supported joint engagement followed by object engagement with little time spent in the other engagement states, including coordinated joint engagement at both time points (see Table 3). Two children who used coordinated joint engagement at Time 1 did not use coordinated joint engagement at Time 2. In addition, four children with DS never used coordinated joint engagement at either time point.

3.1.1. Differences in supported and coordinated joint engagement

Paired sample *t*-tests, indicated that young children with DS spent significantly more time in supported joint engagement than coordinated joint engagement at each Time 1 ($t[15] = 8.31$, $p < 0.001$, $d = 2.08$) and Time 2 ($t[15] = 11.96$, $p < 0.001$, $d = 2.99$).

3.2. Relationship between weighted joint engagement and language abilities

3.2.1. Concurrent relationship between weighted joint engagement and language abilities at Time 1

At Time 1, young children with DS who had higher weighted joint engagement had lower expressive language raw scores on the Vineland at Time 1 when controlling for chronological age, $r = -0.70$, $p = 0.005$. No other significant relationships emerged at Time 1.

3.2.2. Concurrent relationship between weighted joint engagement and language abilities at Time 2

At Time 2, young children with DS who had higher weighted joint engagement had higher expressive and receptive language raw scores on the Vineland when controlling for chronological age ($r = 0.52$, $p = 0.06$; $r = 0.79$, $p < 0.001$). No other significant relationships emerged at Time 2.

3.2.3. Predictive relationship of weighted joint engagement at Time 1 to language abilities at Time 2

Young children with DS who had a higher weighted joint engagement at Time 1 had a lower number of words produced at Time 2 when controlling for chronological age at Time 1, $r = -0.58$, $p = 0.03$. No other significant relationships emerged.

4. Discussion

The present study sought to characterize joint engagement in young children with DS and the relationship between joint engagement and language abilities. Our results indicate that children with DS spent more time in supported joint engagement than in any other engagement state. Further, they spent significantly more time in supported than coordinated joint engagement at both time points. This pattern is consistent with patterns observed in neurotypical 12-to-15-month-olds (Bakeman and Adamson, 1984) and 18-month-olds (Adamson et al., 2009), suggesting that children with DS may demonstrate delays in the use of supported and coordinated joint engagement as compared to their chronological age. However, the pattern of more supported than coordinated joint engagement is consistent with one of the previous studies of joint engagement in 12-to-26-month-olds with DS (Legerstee and Weintraub, 1997). Our study also extends this finding, suggesting that this pattern may continue past the child's 3rd birthday. It is important to note that, Lewy and Dawson (1992) found a pattern of more coordinated joint engagement than neurotypical peers. The age range in this study was quite large, 20–68 months (mean age 37 months). Thus, it is possible that the shift to using more coordinated joint engagement starts after 3 years and increases as children with DS develop. Thus, one important consideration for future research is to examine when children with DS transition to using more coordinated joint engagement than supported joint engagement. This would provide important information about the consolidation of these skills and lead to an increase in reciprocal interactions with others.

In the current study, we used a weighted joint engagement variable to examine the relationship between joint engagement and language abilities at two-time points. At Time 1, children with DS who had higher joint engagement had lower expressive language when controlling for chronological age. This suggests that young children with DS may compensate for their difficulties with expressive language by using joint engagement (Jenkins and Ramruttun, 1998). Research on prelinguistic communication in children with language delays and/or intellectual disabilities has noted similar patterns of using these skills to compensate for expressive language delays (Bishop et al., 2000; LeBarton and Iverson, 2017; Bordenave and McCune, 2021). This is further supported by our finding that children who had higher joint engagement at Time 1 had a lower number of words produced at Time 2. That is, the increased use of joint engagement at Time 1 to compensate for difficulties with expressive language appears to continue into later development (Time 2) as children continue to struggle with expressive language. An alternative explanation for this finding is that caregivers may provide more scaffolding to children with DS who have limited expressive language abilities, leading to an increased frequency of supported joint engagement. This information is particularly important for clinicians to continue to target joint engagement skills, regardless of the strength of these skills in young children with DS, because they are foundational skills that will support later expressive language abilities and outcomes (e.g., Kasari et al., 2008, 2010, 2012).

Nonetheless, it does appear that later in development at Time 2, increased use of joint engagement is associated with higher expressive and receptive language abilities when controlling for chronological age. These findings, that is concurrent relationships at Time 2, align with the view that the dynamic process of joint engagement supports language development (Bakeman and Adamson, 1984; Adamson et al., 2004; Paparella and Kasari, 2004; Bottema-Beutel et al., 2014). Further, the association between joint engagement and receptive and expressive language abilities in DS is echoed in prior research on joint attention—which is similar to joint engagement—and related supportive skills, like triadic eye gaze, (Mundy et al., 1995; Harris et al., 1996; Seager et al., 2018; Hahn et al., 2019). Thus, targeting joint engagement may be especially helpful for language interventions for children with DS into the second year. These interventions may be particularly useful for children with DS who are compensating for expressive language delays with their joint engagement abilities. In addition, it may be especially important to focus on the transition to coordinated joint engagement, which may lead to more salient opportunities for word learning (Mattie and Hadley, 2021) and have a greater impact on language development (Adamson et al., 2004).

Although, the existing interventions that target joint engagement and other prelinguistic communication skills have demonstrated lasting effects on language outcomes for children with intellectual and developmental disabilities and children with language delays (Yoder and Warren, 2002; Fey et al., 2006, 2013; Landry et al., 2008; Kasari et al., 2012), intervention studies that have included children with DS within their samples of children with intellectual and developmental disabilities, have reported mixed findings on their impact for children with DS. For example, Yoder and Warren (2002) found that children with DS had a greater increase in requesting when they were in the no-treatment control group. In contrast, Fey et al. (2006) reported no differences in outcomes if children had DS or not. Nonetheless, our results highlight the importance of teaching caregivers to be responsive to their child's attention when interacting. For example, if a child is engaged with a cat figurine, a caregiver can join their attentional focus by pointing to the figurine saying, "That's a cat." Thus, providing clear linguistic input that can help with word learning (Rowe and Snow, 2020; Mattie and Hadley, 2021). This can also set up an opportunity for the child to respond to the caregiver's communication; thus providing scaffolding that can support the child in moving into supported and coordinated joint engagement. Continued research on the implementation of early language interventions for young children with DS, including caregiver language input, is needed.

4.1. Limitations and future directions

There are several limitations to the present study. First, our sample size is small and a sample of convenience. Although this sample size is not uncommon in DS research, our results should be replicated with larger samples. Also, there was an overlap in chronological ages at Time 1 (12–30 months) and Time 2 (21–38 months). Thus, we were not able to fully explore the ages at which children may start to transition to using coordinated joint engagement with more frequency. In addition, future research should explore if the transition to coordinated joint engagement is facilitated by increased episodes of supported joint engagement with their caregiver at an earlier age. Conducting a more nuanced examination of joint engagement, such as dividing supported joint engagement into lower-order and higher-order skills (Bottema-Beutel et al., 2014) is also needed. Similarly, describing other behaviors associated with joint engagement (i.e., gestures, vocalizations) may also help to elucidate the transition to coordinated joint engagement. It is important to note, that each child had at least 6 months in between their time points and, therefore, reflects changes observed in each child. Therefore, future research is needed to explore age-related changes in joint engagement and its association with language growth.

We also combined data from two pilot studies, as is becoming common practice to achieve larger samples (e.g., shared data repositories), but one of these studies was interrupted by the COVID-19 pandemic leading to a shift in how the data was collected (i.e., *via* Zoom instead of in-person). These different methods may influence how mothers interacted with their child. Although, both involve them being observed by the research team, being at a distance versus physically present in their home may have changed their behavior. However, other than if the family participated remotely, the inclusion criteria and methodology were the same (i.e., instructions for free play, etc.). In addition, the COVID-19 pandemic affected our ability to collect a direct measure of nonverbal cognitive abilities. Future studies should explore the role of nonverbal cognitive abilities on the relationship between joint engagement and language abilities. Person engagement was rarely used in this study. Although, mothers were told to play as they normally would, providing them with a set of toys may have led them to play more with toys than with face-to-face interaction games, like peek-a-boo. We focused our analysis on words produced on the CDI, but this variable does not account for the child's use of sign language. Sign language is often used by children with Down syndrome as a form of alternative and augmentative communication (AAC; Launonen, 1996; Wright et al., 2013). Future studies should also examine the relationship between joint engagement and expressive language as measured by the use of sign language and other forms of AAC. Lastly, both studies focused on early language development. This may have led to more participation from families who were concerned about their child's language abilities.

5. Conclusion

Joint engagement appears to be an important skill for language development in DS. Together these results highlight the importance of teaching caregivers to be responsive during interactions with their child to move them into both supported and coordinated engagement, which in turn may foster language development. Continuing to explore the early language profile, and skills that support it, in DS can help to identify targets for early language interventions in this population. In addition, exploring this profile can help determine the roots of the language difficulties in later development in DS and support the identification of skills that can be targeted early to promote better language outcomes later in development.

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 Illinois at Urbana-Champaign Institutional Review Board. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

LM conceptualized the study, performed the analysis, and drafted and edited the manuscript. DF oversaw data coding and assisted with drafting and editing the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Shared book reading as a context for language intervention for children with Down syndrome: a mini-review

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Acquiring language and communication skills is one of the biggest challenges for children with Down syndrome (DS). However, few evidence-based interventions exist to enhance the development of language and communication in this population. Shared book reading (SBR) is well-established as an effective intervention for language and communication development of typically developing children, and evidence of the possible effectiveness of this approach for those at risk of language difficulties is emerging. This paper provides a mini-review of the existing evidence for SBR in relation to language and communication outcomes for young children with DS. A systematic literature search was conducted with the following inclusion criteria: children with DS aged 0–6;11years, SBR, language or communication outcomes. The results show that interventions which incorporate SBR strategies are associated with improved language and communication outcomes for young children with DS, improved parental sensitivity, and continuing implementation of SBR strategies following intervention instruction. However, evidence is limited in scope, of low quality, including mostly single case studies, with only one study having a control group. We conclude that although SBR may hold promise as a possible intervention, further research is essential to establish what specific components of SBR intervention are most effective for young children with DS and what further adaptations are needed to accommodate the cognitive profile and variability within this population.

KEYWORDS

Down syndrome, intervention, language, communication, shared book reading

1. Introduction

Down syndrome (DS) results from an extra copy of chromosome 21 and is the most common genetic cause of learning disability (Chapman and Hesketh, 2000), affecting approximately 1 in 700 live births (Martin et al., 2009). Language is often one of the biggest challenges for individuals with DS, which can sometimes be below levels expected of non-verbal mental ability (Miller, 1999). Acquiring language is often slow, with expressive vocabulary and grammar being particularly delayed (Abbeduto et al., 2007). Language ability in early childhood is a well-known predictor of later psychosocial and academic outcomes (Snowling et al., 2006), including literacy (Burgoyne et al., 2012; Hulme et al., 2012), hence providing children with DS the opportunity to advance their language skills in early development is crucial to optimize communication, educational, social and wellbeing outcomes.

Although DS is known to present with challenges with language development, few evidence-based interventions are available (O'Toole et al., 2018; Smith et al., 2020). Identifying the most effective way of involving parents/caregivers in supporting achievement of language/communication goals has been identified by the Royal College of Speech and Language Therapists as a key research priority for those with learning disabilities (RCSLT, 2019). Language interventions need to be relevant for the child's social context and easy to implement by parents/caregivers, who are best placed to support their children's language (Roberts et al., 2019). An intervention which is child-centered, relevant for the social context and can be delivered by parents is shared book reading.

1.1. Shared book reading interventions

Shared book reading (SBR) interventions build upon a natural sociocultural activity and focus on augmenting the interaction between the adult and child by using interactive book-sharing strategies, prompts and questioning (Whitehurst et al., 1988). SBR strategies include CROWD ("completion, recall, open-ended questions, wh-questions, and distancing") questions, PEER ("prompt, evaluate, expand, repeat") strategies (Whitehurst et al., 1994), and RAA (Read-Ask-Answer) strategies (Kent-Walsh et al., 2010). The PEER strategy has been adapted to address the needs of children with intellectual disability by adding the "extend" step (PEEER) and provide further prompts. For the purposes of this paper, we will use the term 'SBR' to encompass all approaches.

There is well-established evidence that SBR improves parental linguistic input, and language and pre-literacy outcomes for typically developing children and children at risk of language delay (Huebner and Payne, 2010; Aram et al., 2013; Law et al., 2018). A systematic review of 23 studies by Towson et al. (2021) examined the evidence-base for language outcomes related to SBR interventions for children with language disorder, autism, cerebral palsy, developmental delay and DS ($n = 641$, child age: 35–74 months). A range of effect sizes was reported for expressive (0.44–1.25) and receptive (0.02–1.87) language outcomes, with an overall conclusion of positive improvement and potential for SBR interventions to enhance language outcomes. Another systematic review and meta-analysis by Dowdall et al. (2020), including 19 randomized controlled trials ($n = 2{,}594$) targeting children aged 12–72 months with different language abilities, found that SBR interventions with more than 60 min of total intervention time yielded larger effect sizes for child language outcomes ($d = 0.54$ for expressive and $d = 0.34$ for receptive language) than those of less than 60 min ($d = 0.41$ for expressive and $d = 0.26$ for receptive language). A large effect size for caregiver competence in delivering SBR intervention was also reported ($d = 1.01$).

1.2. Shared book reading and Down syndrome

Whilst some studies focusing on children with developmental disabilities have included children with DS, there is to date no clear synthesis of evidence for the impact of SBR on the language skills of young children with DS. Preliminary evidence suggests that parent–child SBR interactions may be different for parents/careers and children with DS. Parents of 22 children with DS aged 22–63 months used more questions, signs, labels and grammatically simple utterances when sharing a book compared with chronologically age-matched neurotypical children. Children with DS used more nonword vocalizations and gestures, and fewer verbalizations (Barton-Hulsey et al., 2020). Similarly, children with DS have been reported to take a more passive role during reading activities when compared to their peers (van Heerden and Kritzinger, 2008; Al Otaiba et al., 2009). Given the specific behavioral profile associated with DS with a characteristic pattern of strengths and weaknesses (Fidler, 2009) there is a need to evaluate the effectiveness of SBR interventions for this particular group.

Cross-sectional studies of children with DS provide evidence for the ecological validity of SBR interventions. A study of 107 parents/careers of children with DS under the age of 7 years in the United States found that 79% had over 50 books at home and almost all read to their child daily for 10–30 min (Al Otaiba et al., 2009). Based on a survey completed by 191 parents of 1–6 year old children with DS in Ireland, Lusby and Heinz (2020) reported that most parents regularly shared books with their child, and were motivated to do so by social/emotional factors and speech and language development. Parents reported using oral language and print-referencing strategies when sharing books, but also reported challenges in engaging their child in SBR interactions and the need for guidance to enable them to support their child more effectively.

This mini review systematically synthesizes the existing evidence-base for SBR in enhancing language and communication outcomes for young children with DS aged 0–6;11 years.

2. Methods

A systematic literature search was conducted in January 2023 using five electronic databases (MEDLINE via PubMed, PsycINFO, Web of Science Core Collection, ERIC, Cochrane Library). The following search terms were used: [(Down syndrome OR DS OR "trisomy 21" OR disability OR Down's syndrome) AND ("shared book read*" OR "dialogic read*" OR "interactive book read*" OR "book shar*" OR "storybook read*")] which yielded 175 studies after removing duplicates. Titles and abstracts were independently screened for eligibility, according to the following inclusion criteria:

- Study reported results for children with DS aged between 0;0 and 6;11
- Interactive SBR included as part of the study
- Outcomes included at least one child language or communication measure (vocabulary, morphosyntax, communication)
- Published in peer-reviewed journal, in English

Our search identified one hundred and seventy-five studies after duplicates were removed. Of these, one hundred and fifty-five were excluded, twenty were read in full, and of these, seven met the criteria for inclusion. One study was identified through hand-searching of reference lists of the included papers (see Figure 1). From each eligible study, the following data were extracted: participant number, age and

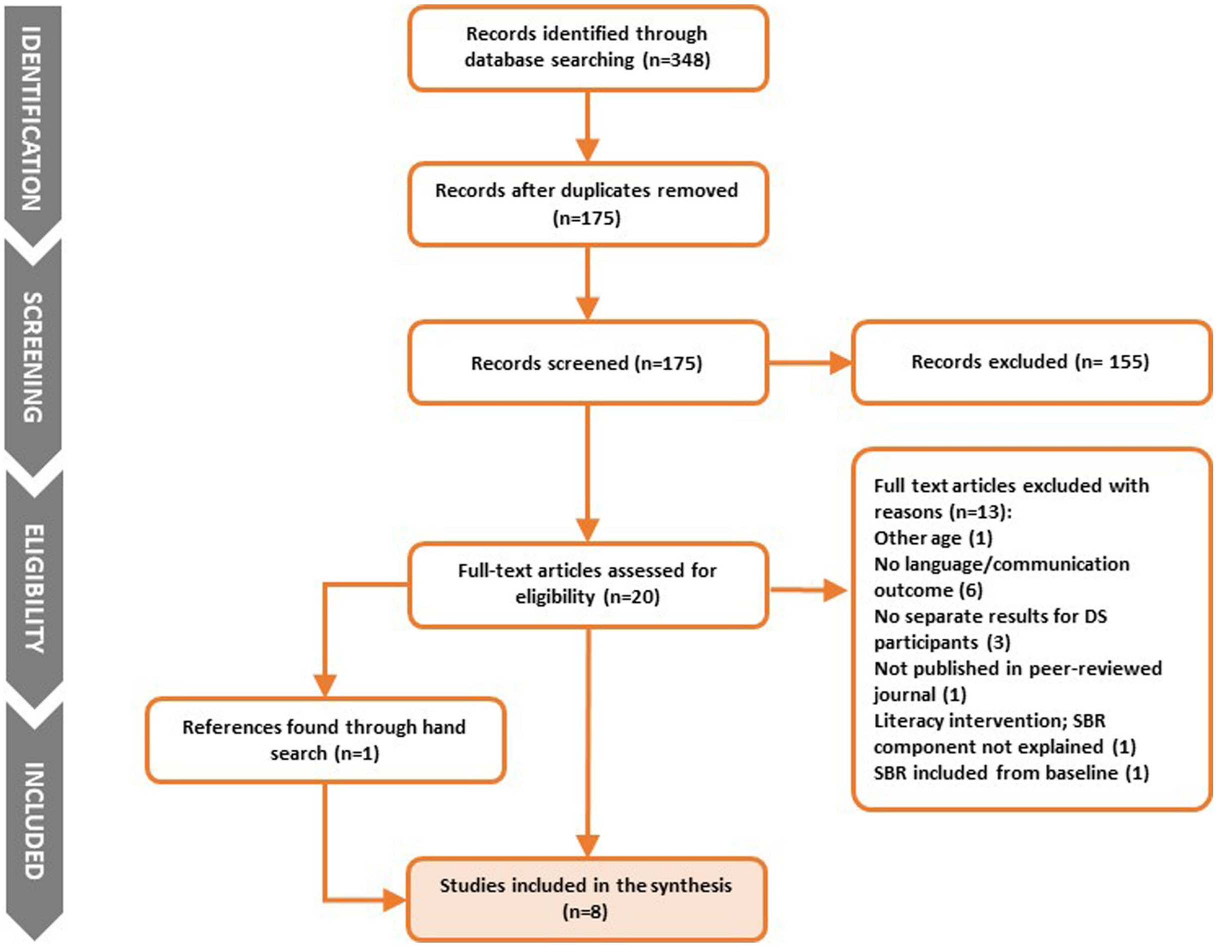

FIGURE 1
Preferred reporting items in systematic reviews and meta-analyses (PRISMA) flow diagram of the study selection procedure.

sex, study design, intervention or material modification details, study aims, parental and child outcomes, and main findings and results.

3. Results

Studies are summarized in Supplementary Table 1 and include five intervention studies (one SBR intervention and four combined interventions which included SBR), two experimental studies, and one observational study. Sample sizes ranged from 1 to 103 children with DS. The ages of participants ranged from 18 to 83 months.

The intervention studies included one randomized-controlled trial including a non-intervention control group (Naess et al., 2022), and four single case experimental designs (Kent-Walsh et al., 2010; Na and Wilkinson, 2019; Pierson et al., 2021; Timpe et al., 2021).

3.1. SBR intervention studies

Pierson et al. (2021) investigated a SBR reading intervention using a case series of four single case studies of children with developmental disabilities, including one child with DS (aged 6 years; 1 month). Caregivers received an initial one-hour training session and weekly one-hour coaching sessions (number not specified) during the intervention phase via video calls, focused on CROWD questions, PEEER strategies, and strategies to support child engagement. The parent delivered three to four reading sessions (of various length) per week to their child totaling 32 sessions. There were no significant changes in the child language outcomes as measured by correct responses to book-related questions except for an increase in the child's comprehension of prompted questions which persisted 1 week after the intervention. There was, however, a significant increase in parental implementation of SBR strategies (see Supplementary Table 1).

3.2. Combined interventions including SBR

Three studies reported interventions that included SBR alongside Alternative Augmented Communication (AAC) interventions. Kent-Walsh et al. (2010) and Timpe et al. (2021) used ImPAACT (Improving Partner Applications of Augmentative Communication Techniques) in conjunction with SBR strategies. Na and Wilkinson (2019) used aided AAC modelling with a 'Strategies for Talking about Emotions as PartnerS' (STEPS) program within the context of book reading where parents asked questions (e.g., what, how, and why) while modelling communication about emotions. Naess et al. (2022) introduced a novel digital vocabulary intervention "Down Syndrome LanguagePlus" (DSL+) using bespoke wordless picture books with video, animation, sounds and voices. They also devised teacher manuals with scripted questions and prompts to encourage literal and inferential talk. SBR activities were combined with structured group tasks to support generalization.

The number of participants with DS ranged from one child (Kent-Walsh et al., 2010; Na and Wilkinson, 2019), three children (Timpe et al., 2021), to 103 children (Naess et al., 2022). Children were aged between 3;0 (Timpe et al., 2021) and 6;11 years (Naess et al., 2022).

Three studies involved parent-mediated interventions delivered one-to-one within the home setting (Kent-Walsh et al., 2010; Na and Wilkinson, 2019; Timpe et al., 2021). One study was classroom-based, delivered by teaching staff (Naess et al., 2022) with a combination of one-to-one, group and whole classroom sessions. Parents and teachers received training ranging from a single one-hour instructional session followed by five sessions with prompting and feedback from the clinician (Na and Wilkinson, 2019) to several hours of in-person and/or online interactive training and continuous support throughout the intervention, including the intervention materials (Timpe et al., 2021) and an intervention manual (Naess et al., 2022). The children received between 11 (Na and Wilkinson, 2019) and 75 intervention sessions (Naess et al., 2022) in total, with the story reading component often lasting about 10 minutes (Kent-Walsh et al., 2010; Timpe et al., 2021; Naess et al., 2022), and ranging between six (Na and Wilkinson, 2019) and 47 minutes (Kent-Walsh et al., 2010). The sessions were spread over a period lasting between three (Timpe et al., 2021) and 15 weeks (Naess et al., 2022). The frequency of sessions ranged from two to three times a week (Kent-Walsh et al., 2010; Na and Wilkinson, 2019; Timpe et al., 2021) to daily sessions (Naess et al., 2022).

Regarding child language and communication outcomes, Naess et al. (2022) found a significant intervention effect for trained vocabulary immediately post-intervention compared to non-intervention controls but there were no group differences on standardized vocabulary or grammar measures. Timpe et al. (2021) reported an increase in the frequency of communicative turns and novel semantic concepts recorded during reading activities post-intervention. Na and Wilkinson (2019) reported an increased number of child utterances related to the communication of emotions post-intervention, which was maintained during the generalization phase and at follow-up, 2–6 weeks later. Kent-Walsh et al. (2010) reported an increase in the total number of communicative turns and semantic concepts used post intervention which were maintained for several weeks (see Supplementary Table 1).

Parent outcomes were reported by three studies: increased accuracy in parental use of the RAA strategy post-intervention compared with baseline (Timpe et al., 2021), increase in number of open-ended questions used by the parent (Na and Wilkinson, 2019) and increase in use of communication partner interaction strategies by the parent (Kent-Walsh et al., 2010) – see Supplementary Table 1.

3.3. Non-intervention SBR studies

Three non-intervention studies used experimental (Burgoyne and Cain, 2022; Frizelle et al., 2022) or observational (Hilvert et al., 2022) designs to investigate SBR interactions between children with DS and their parents. The number of participants ranged from 8 to 15. Children were aged between 1;6 and 6;9 years.

Two studies adapted SBR materials to address the needs of children with DS. Burgoyne and Cain (2022) embedded 12 questions within a book to support parents to ask questions about literal and inferential information. Frizelle et al. (2022) embedded key-word signing within books to encourage child participation (signed condition) and compared it to reading a book as usual (unsigned condition). Hilvert et al. (2022) investigated the differences between maternal and paternal language input during SBR.

Differences in child language were observed in both experimental studies. Burgoyne and Cain (2022) reported that children with DS produced significantly more utterances, significantly more words and more different words when parents used question prompts compared to the typical reading condition. Frizelle et al. (2022) found that children attempted to sign significantly more in the signed than unsigned condition (see Supplementary Table 1).

Modification of materials encouraged parents to focus more on extra-textual talk (Burgoyne and Cain, 2022) and increased the number of parent utterances (Frizelle et al., 2022). Hilvert et al. (2022) found that mothers produced more utterances and used more descriptive language than fathers, while fathers read significantly more verbatim. Despite these differences, parents spent most of the book reading interaction engaged in contextualized talk (76%), followed by reading (21%), and decontextualized talk (3%) and both mothers and fathers used more complex language with children who had better language skills (see Supplementary Table 1).

4. Discussion

This mini-review contributes towards better understanding of the potential of SBR as a possible intervention for children with DS to enhance language and communication skills. The key findings are that interventions which incorporate SBR are associated with improved language and communication outcomes for young children with DS and that studies involving parents/careers, report changes in adult behavior and language input following the adoption of the SBR strategies. Importantly, parents/careers perceive the intervention as effective, easy to implement and enjoyable. However, the evidence is limited in scope, largely of low quality with only one intervention study including a control group. SBR is often combined with other interventions, making it difficult to identify any unique effects on language outcomes that may be attributable to SBR, but also suggesting that SBR strategies may be beneficial if used in combination with another intervention to enhance children's language and communication skills. Non-intervention experimental and observation studies provide some support for the potential of SBR to enhance language and communication outcomes for children with DS, with evidence of question prompts and the use of key-word signing in SBR being associated with increased child participation and communication. These findings are consistent with findings of previous reviews of SBR with other populations (Mol et al., 2009; Dowdall et al., 2020; Towson et al., 2021). Parents often lack in confidence and seek advice on how to optimize these interactions with their children, and manage their child's attention and engagement (Barton-Hulsey et al., 2020; Lusby and Heinz, 2020). This highlights the need for parent/career support for SBR, and for further research to identify effective ways to enable parents to support their child's attention, behavior and cognitive needs during SBR activities.

Expressive language is typically an area of relative weakness in children with DS, compared with receptive language (Seager et al., 2022). This mini-review identifies increases in children's expressive language following SBR (Na and Wilkinson, 2019; Timpe et al., 2021; Naess et al., 2022) which is also supported by existing reviews (Dowdall et al., 2020; Towson et al., 2021). This could be because SBR strategies aim to encourage children to take an active communicative role, and provide opportunities for parents to model and scaffold language in a naturally occurring context (Mol et al., 2008; Towson et al., 2021; Burgoyne and Cain, 2022). Previous studies report large effect size ranges for language outcomes which could be due to different research designs and/or measures used; this further suggests the need for future research to establish which SBR components promote improvement in language outcomes for different populations (Dowdall et al., 2020; Towson et al., 2021).

This review shows that SBR strategies have been implemented through the instruction of parents/careers/educators which can lead to behavior modification in the adult and this in turn can have an effect on the language and communication outcomes of the children with DS. This suggests effective implementation within the child's natural environments, thus emphasizing the potential for SBR strategies to generalize beyond the intervention sessions. Involving parents/careers is essential to enable the creation of a child and family-centered intervention (Alsem et al., 2017) and SBR naturally lends itself to this approach. It should be noted that parental input may vary between mothers and fathers during SBR (Hilvert et al., 2022), and that parents adapt their language according to their child's chronological age and language ability (Lusby and Heinz, 2020; Hilvert et al., 2022). This needs to be further explored with more controlled studies examining the possible relation between differences in parental input during SBR and child language outcomes.

Given the cognitive profile and variability that exists within the DS population (Onnivello et al., 2022), it is possible that some children may need different levels or types of adult support, specific dosage or implementation adaptations (Burgoyne and Cain, 2022). Other reviews have identified incomplete reporting of child and adult demographics including ethnicity and home language, child intellectual abilities and additional diagnosis to be the limiting factors when synthesizing effectiveness of SBR interventions (Dowdall et al., 2020; Towson et al., 2021). Burgoyne and Cain (2022) found considerable variability in parent shared reading behaviors and child engagement. They note a case of a younger child who spent less time engaging in extra-textual talk and produced less language when sharing a book with embedded prompts. This was in contrast with the behavior noted in the older children who engaged better and produced more language when parents made reading more interactive. This suggests that SBR strategies may need to be modified and adapted for children of different ages and/or attention and language skills to engage with SBR. Small-scale research has suggested that incorporating pause time (Towson et al., 2021), pictures (Whalon et al., 2013), prompts (Burgoyne and Cain, 2022) and technology enhancement (Grygas Coogle et al., 2018; Quinn et al., 2020; Naess et al., 2022) may be effective strategies in SBR with children with developmental disabilities. Moreover, interventions included in this mini-review were of variable dosage (between 11 and 75 intervention sessions in total) and dosage has been found to mediate SBR intervention effectiveness (Dowdall et al., 2020). However, due to the heterogeneity of the reported outcomes, the variability of the measures used and the fact that few studies reported actual effect sizes (see Supplementary Table 1), it is difficult to estimate for our set of studies whether dosage may have mediated the effectiveness of SBR interventions. Future research should consider the optimum dosage of intervention, which may vary among different groups. Furthermore, most studies included here measured outcomes during, or immediately after, the intervention. This lack of longer-term follow-up results means that evidence of lasting effects is currently missing and future research should bridge this gap to inform SBR practices for children with DS.

Although it is difficult to draw definitive conclusions based on the limited available evidence, the studies included in this mini-review suggest that SBR is a promising intervention approach which could be implemented with children with DS to enhance their language and communication skills.

Author contributions

MJ, EP, VS, and KB contributed to the conception and design of the study. MJ conducted the literature searches and data extraction. EP and MJ wrote the first draft of the manuscript. All authors contributed to manuscript, read, and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2023.1176218/full#supplementary-material

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The impact of COVID-19 (Coronavirus) on children and young people with Down syndrome in the United Kingdom

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The COVID-19 pandemic had a profound impact across the globe. Evidence suggests children with Special Educational Needs and Disabilities and their families experienced impacts on well-being and disruptions in support from education and health services. This study investigated the impact of measures associated with the COVID-19 pandemic on children and young people (CYP) with Down syndrome in the United Kingdom, specifically changes in speech, language and communication abilities, behavior, social, emotional and mental health and access to education and healthcare services. Forty-six parents/carers of CYP with Down Syndrome (aged 2–25 years) completed an online survey between June and September 2020. Parents/carers frequently reported deterioration in speech, language and communication, literacy and attention skills since the onset of the pandemic. Deterioration in social and emotional wellbeing and behavior, including greater reliance on adults were also reported for some CYP with Down syndrome. Parents reported challenges with home-schooling and reductions in support from education and community services. Preferences for support during COVID-19 were for professional support or from other parents. These findings have implications for the support that is now needed for CYP with Down syndrome and their families and for periods of social restrictions in the future.

KEYWORDS

Down syndrome, COVID-19, mental health, communication, education, healthcare, speech and language therapy

1. Introduction

The COVID-19 pandemic has had a profound impact across the globe, including increased levels of anxiety and stress in the population (Salari et al., 2020; Shevlin et al., 2020) and consequences for the social and emotional wellbeing of children and young people (CYP) (Ma et al., 2021; Racine et al., 2021). In the United Kingdom, measures were introduced from March 2020 resulting in school closures and national and regional lockdowns, reducing time spent outside of home and interactions with others outside of immediate families. These measures continued until December 2021 (Institute for Government Analysis, 2022).

Evidence suggests that CYP with Special Educational Needs and Disabilities (SEND, an accepted term used in education in the United Kingdom) and their families may have been disproportionately affected by COVID-19 measures. In data collected early in the first

United Kingdom lockdown (March–April 2020) parents of children with SEND reported raised levels of anxiety, stress and low mood in both themselves and their children, in addition to fears and worries specifically related to their child's SEND (Asbury et al., 2021). Data collected at a similar time provides further evidence to suggest parents/carers of CYP with SEND were directly affected by COVID-19 measures. A survey of 415 parents/carers carried out in April 2020 in a region of England reported by O'Hagan and Kingdom (2020) found that 65% were physically and mentally exhausted, and 51% reported increased anxiety and depression. Subsequently, Sideropoulos et al. (2022a) compared parental report of anxiety and worries for their child with SEND ($n = 407$) and their neurotypical siblings to examine how COVID-19 measures affected young people with SEND differently. Data in relation to individuals across a wide age-range (1–45 years) including a range of diagnoses and a high prevalence of intellectual disabilities (76%), indicated raised anxiety in children with SEND in the first few months of the pandemic, as compared with their siblings. Awareness of COVID-19 and parents with greater anxiety predicted higher reported anxiety levels for individuals with SEND.

Families have also been impacted by changes and/or interruptions in their access to education and health services. Children and young people with SEND typically access support from multiple services, many of which were disrupted by the pandemic (Jeste et al., 2020). Major changes in the delivery of community children and young people's services were reported, in part due to re-deployment of staff to support an urgent pandemic response (Chadd et al., 2021; NHS Confederation and NHS Providers, 2022). For example, many children received reduced speech and language therapy during lockdown, putting them at increased risk of poor academic attainment, and difficulties with friendships, social functions and mental health (Royal College of Speech and Language Therapists, 2021). Collating the data from three United Kingdom surveys of almost 5,500 parents/carers with children with SEND, O'Hagan and Kingdom (2020) reported the following themes: (1) reduction in external support from schools and therapy services, (2) parents and neurotypical siblings providing more care to their child/sibling with SEND, and (3) difficulties managing home learning due to school closures. Toseeb et al. (2020) conducted a survey of 339 parents/carers of CYP (aged 5–18 years) with SEND between March and May 2020 and found that less than half of parents of children with SEND reported that the support they received during the first lockdown was sufficient to meet their child's needs. Interestingly however, there is also some evidence that school closures and lockdowns may have been associated with positive impacts for some children with SEND. In a survey carried out in June 2020 with ~1000 parents/carers, 38% reported an improvement in their child's anxiety during the first lockdown, with reasons for this including less pressure, a less formal learning environment at home, better understanding of the child's needs and fewer sensory issues (Special Needs Jungle, 2020).

Down syndrome is a common cause of SEND, with a prevalence of 25.4 per 10,000 total births in England (National Congenital Anomaly and Rare Disease Registration Service, 2019) and a population prevalence estimated at 10,438 children aged 0–18 years in England and Wales (Wu and Morris, 2013). Down syndrome is associated with a specific cognitive profile, including intellectual disability, and difficulties related to speech, language and communication, attention and executive function, with the level of need increasing as children reach school-age (Grieco et al., 2015). Relative strengths have been observed in receptive language and social use of communication (relative to mental age) with particular needs in the domains of expressive language (grammar and syntax) and phonology (Abbeduto et al., 2007; Laws et al., 2015). Down syndrome is also associated with a specific profile of literacy skills, and while strengths in reading and in particular word identification skills have been reported, there is also evidence that CYP with Down syndrome have difficulties with phonological awareness and decoding (Snowling et al., 2008; Loveall and Barton-Hulsey, 2021). There is however variability in the strengths and needs of children with Down syndrome. A recent exploratory study of 72 children reported multiple cognitive profiles: (1) similar levels of verbal and nonverbal abilities, (2) specific needs in the verbal domain relative to non-verbal abilities, and conversely: (3) more profound needs in nonverbal abilities as compared with verbal abilities (Onnivello et al., 2022). Children with Down syndrome are also reported to have a specific profile related to social, emotional and behavioral functioning. There is some evidence from a study of 8-year-olds that children with Down syndrome have lower levels of anxiety, when compared with data from 8-year-olds in the general population (Van Gameren-Oosterom et al., 2011). Strengths have been reported in pro-social behaviors alongside a higher risk of externalizing behaviors at school-age (e.g., impulsivity, disruptive behaviors) and higher rates of internalizing difficulties emerging later such as anxiety, depression and withdrawal, relative to other children with intellectual disability (Grieco et al., 2015).

There has been limited research focused on the impact of COVID-19 on CYP with Down syndrome specifically. Studies of CYP with SEND have included a broad range of different diagnoses, and while parents/carers of CYP with Down syndrome were included in some studies, these amounted to less than 3% of the total sample (less than 10 parents/carers; Toseeb et al., 2020; Asbury et al., 2021). A larger number of parents/carers of children with Down syndrome were recruited by Sideropoulos et al. (2022a) (n=103, 26% of the total sample) but no syndrome-specific conclusions can be drawn from these findings due to the heterogenous nature of the sample in terms of types of SEND. Data from a sample of adults with Down syndrome in Italy taken before the pandemic and during lockdown measures suggests a range of impacts, including a decrease in independence, increase in social withdrawal, decrease in aggressive behavior, and an increase in depressive symptoms (Villani et al., 2020). To our knowledge, only one study has reported on the impact of COVID-19 on CYP with Down syndrome. Sideropoulos et al. (2022b) compared data from 115 caregivers of CYP with Down syndrome with data from caregivers of children with different types of SEND before the pandemic, during the first lockdown (March 2020) and in January–March 2021 in the United Kingdom. Reported anxiety levels were higher during the pandemic than pre-pandemic levels for both groups, but lower overall for participants with Down syndrome. Awareness of COVID-19, health problems and diagnoses of anxiety disorder were predictors of anxiety for participants with Down syndrome and with other types of SEND.

This study aimed to extend findings reported previously by investigating the impact of measures associated with the COVID-19 pandemic on CYP with Down syndrome in the United Kingdom, specifically investigating changes in speech, language and communication abilities, behavior, social and emotional functioning, and mental health and access to education and healthcare services.

2. Materials and methods

2.1. Participants

Parents/caregivers of 151 children and young people (CYP) aged 2 to 25 years with speech, language and communication needs (59% male, mean age 9.9 years ranging from 2 to 25 years) from across the United Kingdom were recruited through parent networks and support groups (e.g., Mumsnet, Facebook groups, university parent and family networks), speech and language therapy networks and social media as part of a larger study investigating the impact of COVID-19 on CYP with speech, language and communication needs. No incentives were offered for participation. For the purposes of this study, participants were included if their child had a diagnosis of Down syndrome ($n = 50$). Four respondents were from the United States and were excluded.

2.2. Materials and procedure

Parents/caregivers completed an anonymous online survey via Qualtrics between 26th June and 31st September 2020. This followed the first United Kingdom lockdown, which was from 23rd March 2020 to the end of June 2020. The survey included a combination of multiple choice, Likert scale and open-ended questions in 5 sections: (1) demographic information about the parent/caregiver and the child (12 questions), (2) information about the child's SEND, speech, language and communication needs and specialist support in place (4 questions), (3) support from education and healthcare services since the onset of the COVID-19 pandemic (5 questions), (4) impact of COVID-19 on children's speech, language and communication abilities, social, emotional and behavioral functioning (10 questions), and (5) impact of COVID-19 on access to education and healthcare services and support needs (5 questions). Two further questions were included to check attention and appropriate responses to mitigate for fraudulent responses (Q13 and Q23, see Supplementary materials). The survey took approximately 20–30 min to complete, and parents had a week to complete the survey after starting (see Supplementary materials for the full survey).

The survey was piloted with five parents/caregivers of children with SEND and five professionals working with children and young people with SEND. Overall, participants reported they understood the questions and felt the length of the survey was appropriate but remarked that the sequencing of questions did not always flow logically. In response to this feedback, the questions were re-ordered and grouped into themes. Clarification was also requested for a few questions, and these were then rephrased to make them clearer on first reading.

2.3. Ethics statement

Ethical approval was granted by the Faculty of Science and Health Ethics Sub-Committee at the University of Essex before the start of the study. Respondents completed online consent prior to completing the survey and were reminded that their participation was voluntary and they were free to withdraw from the study at any time. Consent was required in order to proceed with the questionnaire.

2.4. Data analysis

2.4.1. Analysis plan

The data collected was predominantly quantitative. Qualtrics, the online survey platform was used to host the questionnaire. Inbuilt into the platform is bot detection software which was enabled to identify multiple responses, and the settings were also changed to prevent multiple submissions from one respondent in addition to monitoring respondent metadata to determine if the same respondent was completing the survey on multiple occasions. The survey responses were then inputted to IBM SPSS Statistics for Windows, Version 27.0 (IBM Corp, 2020). A missing value analysis identified any additional inconsistencies and ensured that data categories were re-labelled where appropriate (i.e., changes from ordinal to nominal).

As the study was investigating the impact of COVID-19 on (a) speech, language, communication, behavior, social and emotional functioning, and (b) access to education and healthcare services for United Kingdom-based children and young people with Down syndrome the survey questions were designed to explore each of these aspects in detail. The analysis plan then centered around addressing these different domains by descriptively analyzing the proportions of respondent answers. The survey included free text boxes where, (a) respondents could write additional information if the options listed did not cover their experience, or (b) to give additional detail to a previous question. For example, 'Was it more difficult to book health/well-being appointments since the onset of COVID-19?', a yes/no answer, was followed up with a question asking *why* it was more difficult if respondents had answered 'yes'. This served to give more detail and nuance to the set survey answers, but were not in-depth responses as characterized by qualitative research. Consequently, qualitative methods such as narrative analysis, discourse analysis or grounded theory were excluded in favor of a content analysis approach more suited to short form answers. The qualitative content analysis then supplemented the quantitative data, which answered the principal research questions. The content analysis plan was approached thus: Systematic reading of all free text responses per survey question; Formulation and quantification of nature of responses; Weighting of responses by frequency of use; Construction of themes and cross-checking with the research team; Discussion of findings, noting both commonalities in response and outlying experiences.

2.4.2. Data analysis

Quantitative responses were analyzed using descriptive statistics including frequency information. Open-ended responses were analyzed using content analysis. Content analysis was used as a way to 'organize and elicit meaning' from the free text responses (Bengtsson, 2016), and to provide more detail and depth to the predominantly quantitative survey data collected as part of this study. A manifest analysis approach was employed, where researchers describe what respondents say and quote verbatim, as opposed to the more interpretative latent analysis, where the underlying meaning of a text is sought (Bengtsson, 2016). The authors were all involved in the content analysis process; as different stages were completed, results were discussed in context of the survey findings. Key themes arising from the analysis are reported, along with less dominant, but nevertheless notable insights.

3. Results

3.1. Demographic information

Forty-three participants (93%) were mothers (1 father, 1 grandparent, 1 other) and 43 (93%) were of white ethnic background (2 mixed race, 1 Chinese/Chinese British). Twenty-four (52%) were employed. Total household income ranged from less than £16,000 to more than £120,000. Ten (22%) participants were from households with a total income below the poverty threshold for 2020–21 (Joseph Rowntree Foundation, 2023).

The mean age of the CYP with Down syndrome was 10.2 years (range 2–25 years: nine aged 2–5 years; eighteen aged 6–11 years; twelve aged 12–18 years; three aged 19–25, and; four undisclosed). Twenty-three (50%) of the CYP with Down syndrome were male. Forty-two (91%) of the CYP were of white ethnic background (3 mixed race, 1 other) and 37 (80%) had siblings. Families responded from all regions of England ($n = 32$, 70%), Northern Ireland ($n = 8$, 17%), Ireland ($n = 3$, 6.5%) and Wales ($n = 3$, 6.5%). Five families (11%) reported speaking a language other than English at home (3 Spanish, 1 Afrikaans, 1 Portuguese).

Twenty-seven (59%) of the children were reported to have a diagnosis of Down syndrome only, with the remaining 19 (41%) having multiple SEND diagnoses (see Table 1). Twelve parents reported their child had a diagnosis of learning disabilities (26%) and 17 reported some form of sensory impairment (37%). There was a high prevalence of pre-existing speech, language and communication needs reported (present prior to the pandemic). The most common were difficulties with speech sounds ($n = 42$, 91%), expressive language difficulties ($n = 34$, 74%), reading and writing ($n = 29$, 63%), receptive language difficulties ($n = 18$, 39%), and stammering ($n = 10$, 22%). Pre-existing social, emotional and behavioral difficulties were also reported by some parents (see Table 1).

CYP with Down syndrome had a range of educational placements, including nursery ($n = 5$, 11%), mainstream school ($n = 20$, 43%) and specialist school ($n = 15$, 33%) settings (see Table 2). Forty (87%) had an Education, Health and Care Plan (a legal document in the United Kingdom describing a child's SEND and the support needs of the child).

3.2. Awareness of COVID-19

Overall, a relatively low level of awareness of COVID-19 was reported: 33 of the children (79%) were reported to rarely or never ask questions about COVID-19 and only 15 parents/caregivers (36%) agreed that their child was very aware of COVID-19. Parents reported a low frequency of child worries specifically about getting COVID-19 ($n = 4$, 9%) or leaving the house ($n = 2$, 4%). Few parents reported that their child was experiencing fears in this period (losing friendships $n = 5$ 11%, getting ill $n = 3$ 7%, losing parents $n = 3$ 7%, losing grandparents $n = 2$ 4%, dying n = 1 2%). Most parents/caregivers reported that it was difficult to explain COVID-19 to their child ($n = 41$, 89.2%), and a variety of resources were drawn upon to do so - most commonly social stories ($n = 15$, 32.6%), telling stories ($n = 11$, 23.9%), information from teachers/schools ($n = 13$, 28.3%) and their children talking with friends ($n = 9$, 19.6%).

TABLE 1 Co-occurring pre-existing special educational needs and disability diagnoses and difficulties reported by parents.

SEND diagnosis	Frequency	Percent
Hearing impairment	9	19.6
Visual impairment	6	13.0
Autistic spectrum disorder	6	13.0
Developmental language disorder	5	10.9
Moderate learning disability (MLD)	5	10.9
Language disorder	4	8.7
Profound and multiple learning difficulty (PMLD)	4	8.7
Dyspraxia	3	6.5
Severe learning disability (SLD)	3	6.5
Attention deficit hyperactivity disorder	2	4.3
Multi-sensory impairment	2	4.3
Social communication disorder	2	4.3
Other	1	2.2
Speech, language and communication difficulty		
Difficulties with pronunciation/production of speech sounds	42	91.3
Difficulties with talking–using words and sentences	34	73.9
Reading and writing	29	63.0
Difficulties with understanding what other people say	18	39.1
Stammering/stuttering/dysfluency	10	21.7
Voice–affecting the voice box and how the voice sounds	3	6.5
Social, emotional and behavioral difficulty		
Social interaction difficulties	14	30.4
Behavior problems	10	21.7
Emotional difficulties	7	15.2

3.3. Impacts of COVID-19 measures on CYP with Down syndrome

Twenty-eight respondents (67%) reported concerns that their child's communication, learning and development had deteriorated since the onset of the pandemic. Table 3 shows that deterioration in communication, social/emotional skills, attention and learning, play, skills for daily living, and physical health were reported with the most frequently cited areas of deterioration being social skills ($n = 20$, 43.5%), speech sounds ($n = 15$, 32.6%), communication ($n = 14$,

TABLE 2 Educational setting (please note two participants reported more than one educational setting).

Educational setting	Frequency	Percent	Attending school during pandemic	% Attending school
Nursery	5	10.9	1	20.0
Mainstream primary	17	37.0	4	23.5
Mainstream secondary	3	6.5	1	33.3
Special school	14	30.4	5	35.7
Further education college	3	6.5	0	0.0
Specialist independent college	1	2.2	0	0.0
Adult services/community	2	4.3	0	0.0
Home educated	1	2.2	0	0.0
No school place	2	4.3	0	0.0

TABLE 3 Areas of deterioration in children as noticed by parents during the COVID-19 pandemic.

Area of deterioration	Frequency	Percent
Communication		
Communication	14	30.4
Speech/pronunciation/production of speech sounds	15	32.6
Stammering/stuttering/dysfluency	4	8.7
Language/understanding what other people say	6	13.0
Language/talking–using words and sentences	12	26.1
Voice	1	2.2
Social/emotional		
Social skills	20	43.5
Behavior	5	10.9
Emotional wellbeing	10	21.7
Attention and learning		
Attention	13	28.3
Memory	5	10.9
Reading and writing	10	21.7
Play	4	8.7
Skills for daily living	6	13
Physical health		
Physical difficulties impairing their ability to walk and/or move and/or talk	1	2.2
Physical health	1	2.2
Other		
Numeracy	1	2.2

30.4%), attention ($n = 13$, 28.3%), and using words and sentences ($n = 12$, 26.1%). Deterioration in physical skills, voice, play and stammering were rarely reported across the sample. A high level of parental concerns relating to the CYP with Down syndrome were reported, most commonly in social interactions ($n = 40$, 87%), meeting learning/development needs ($n = 38$, 82.6%) child health ($n = 36$, 78.2%), speech, language and communication development ($n = 36$, 78.2%) and emotional wellbeing ($n = 36$, 78.2%).

Ten (24%) parents/carers reported an increase in anxiety in CYP with Down syndrome. Parents/carers conveyed a range of increased or new adverse behaviors in their child since the onset of the pandemic, most frequently increased reliance on television ($n = 22$, 48%), reduced motivation ($n = 21$, 46%), loneliness ($n = 18$, 39%), and greater dependence on adults ($n = 17$, 37%). Parents expanded on their responses in the free text boxes, describing the effects of isolation and anxiety as their children missed their routines, school friends and extended families. A parent of an only child described how her daughter had not seen other children for months curtailing any meet ups with school friends:

> "she doesn't understand social distancing and will hug strangers in public places." (R9: 93)

Others recounted similar experiences, with children missing friends and social activities such as going to the cinema and discos. The lack of social interaction meant that social skills had been forgotten:

> "Speech and language is my child's main challenge but she needs face-to-face support which hasn't been possible […] her need to socialise with other children has been a daily struggle." (R174: 91)
>
> "Her social communication has regressed. Now she is back at school she's currently not interacting with peers." (R234: 90)

A less prominent but nevertheless recurring theme in the free text responses was the positive effects of staying at home during the pandemic. Respondents still expressed concern that their children were falling behind educationally, but reported that their child had benefitted in terms of social skills and prolonged family contact:

> "I fear my son's learning will have regressed significantly, particularly maths. On the positive side his life skills have improved and we have enjoyed lots of time as a family." (R179: 91)

Elsewhere, parents noticed improvements due to increased time interacting with siblings. One respondent noticed their daughter's speech and creative play had developed due to increased one-on-one time with her sister. Another noticed improvements for the same reasons, contrasting it with her daughter feeling pressure to interact with peers at school:

> "[She's] much happier without the stress of having to conform." (R136: 91)

Similarly, a parent/carer reported that their son was:

> "flourishing and taking good learning risks because he's not under the microscope or the 'evaluative gaze' at home." (R150: 91)

Another respondent recounted how their son's school had noticed how much progress he had made during the first lockdown:

> "I put this down to us being at home all day […] so he got established in routines and play activities in a way he hadn't before with typical family busyness." (R200: 90)

In this instance, the parent/carer also reported that the school were posting videos of activities on Twitter and Instagram and via an online learning journal very effectively and supporting parents/carers in setting up appropriate activities at home. In one instance, a parent mentioned paying for tutorials tailored specifically for children with Down syndrome, which was viewed as an excellent resource, stating:

> "[It's] just a shame that we can't look at funding this for all pre-school children with DS, now that location is not a barrier." (R195: 90)

3.4. Impact of COVID-19 measures on access to education

Thirty-five (76%) children had not attended their educational setting during the pandemic. Of those with mainstream placements, 25% ($n = 5$) were attending school, and of those with special school places, 36% ($n = 5$) were attending school (see Table 2). Of the children who were not attending school, 17 (48.6%) parents/caregivers reported their child was frequently missing being at school and 19 (52.3%) reported finding the experience of home-schooling difficult. Seventeen (48.6%) parents reported receiving some support from school to home-school their child. One respondent expressed frustration that the school appeared to have forgotten that her daughter's work needed to be differentiated or tailored to her needs:

> "I just feel that schools not having any work to give pupils that need an element of contact with other pupils was a big let-down and caused social isolation. Group work or a group online session would have helped this." (R151: 91)

This parent concluded that their daughter would not have done any work at all had they not been a trained Teaching Assistant themselves and therefore able to personally provide the specialist support required at home. A very similar experience was recounted by another parent who described how the work given to their son was not differentiated to his level. Another respondent wanted their child's school to do more live online teaching so their child could see his peers rather than short videos which were not easy to engage with.

Parents reflected on the challenges of home working, home schooling and lack of support engendered by the lockdowns:

> "Families who are expected to work from home with children with special needs have been effectively made to neglect their children. It has been a horrendous year for us as a family […] I am now furloughed which has made a big difference, but back at work soon with no childcare or support." (R174: 91)

3.5. Impact of COVID-19 measures on access to community services

Figure 1 shows that many support services stopped, were delivered differently or reduced during the COVID-19 pandemic. All CYP with Down syndrome receiving speech and language therapy experienced a change in delivery in the months prior to completing the questionnaire: 63% ($n = 25$) reported that their speech and language therapy provision was stopped, 30% ($n = 12$) reported it was delivered in a different way and 7% ($n = 3$) reported it was reduced. Eighty-three percent ($n = 20$) of those receiving occupational therapy experienced a change in delivery: 54% ($n = 13$) reported that it had stopped, 17% ($n = 4$) that it was delivered differently and 13% ($n = 3$) that it had reduced. Physiotherapy and clinical psychology were either stopped ($n = 11$, 85% and $n = 3$, 50% respectively) or continued without change ($n = 2$, 15% and $n = 3$, 50% respectively).

Beyond health services, 54% ($n = 13$) of those receiving support from specialist teachers continued to receive this assistance, mostly in a different way. Educational psychology support was either stopped ($n = 4$, 57%), reduced ($n = 1$, 14%) or continued unchanged ($n = 2$, 28%). Of all support services, social work and school counseling/pastoral care were most likely to continue during the COVID-19 pandemic ($n = 9$, 82% and $n = 5$, 71% respectively), although note the small numbers of families accessing these services (see Figure 1).

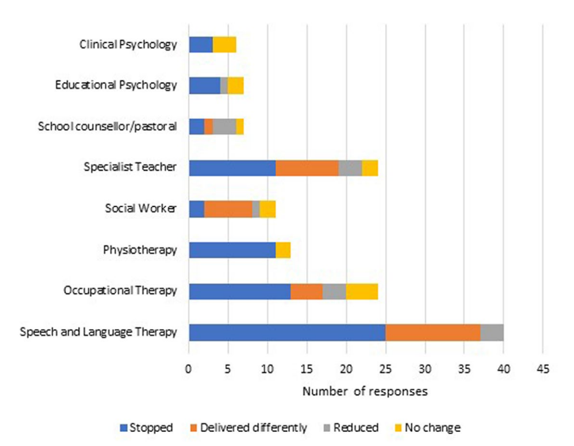

FIGURE 1
Change in delivery of specialist support during the COVID-19 pandemic.

Twenty-seven parents/carers (58.6%) reported finding accessing healthcare/wellbeing-related appointments harder than usual since the onset of the COVID-19 pandemic with reasons given including canceled appointments, and difficulties accessing telehealth. The free text responses revealed that 14 parents/carers had appointments canceled with a range of services including audiology, optometry, dentistry in addition to hospital and clinic appointments. Two parents/carers were able to access health advice over the phone or via an online appointment, for example by speaking to a Community Specialist Health Visitor. Another respondent had organized an online meeting but had problems with their internet connection. One parent/carer was informed that they could not access speech and language therapy online because the service did not have laptops available for videocalls. Some respondents reported care being delayed, for example an annual blood test.

For some, personal circumstances made it very challenging to attend appointments. One parent/carer described how difficult it was to make an appointment because their son is a vulnerable adult; one parent/carer could not attend appointments because their child would not wear personal protective equipment (PPE). Childcare was an issue, with some parent/carers struggling to find childcare for younger siblings. Concerns over catching COVID-19 or shielding prevented further parent/carers from arranging appointments. Another parent had tried unsuccessfully to speak to a GP or pediatrician about their child's COVID risk level. For those who were offered online appointments, respondents reported problems with their children engaging with videocalls.

Overall, 25 of the respondents (60%) reported that they rarely or never received support for their child when they needed it during the pandemic. Parents reported that support regarding their child's learning ($n = 32$, 70%), speech ($n = 23$, 50%), language ($n = 21$, 46%), social skills ($n = 21$, 46%), emotional status ($n = 19$, 41%) and health/physical development ($n = 14$, 30%) would have been helpful. When asked for their preferences regarding support in the context of a pandemic, most parents/carers reported their preference for receiving support and information from a professional ($n = 39$, 85%) and in some cases, support from a professionally trained parent ($n = 15$, 33%) or a community of parents ($n = 15$, 33%). When asked what form they would prefer to receive help, parents stated by videocall ($n = 24$, 52%), online materials ($n = 22$, 48%), WhatsApp/text ($n = 13$, 28%), online videos ($n = 13$, 28%), written materials ($n = 9$, 20%), podcasts ($n = 8$, 17%), and online helplines ($n = 7$, 15%). Few parents selected the following as helpful support tools: a telephone helpline ($n = 4$, 9%) books/pamphlets ($n = 2$, 4%) or via the media ($n = 1$, 2%).

4. Discussion

To our knowledge, this is the first study to report on a wide range of impacts of COVID-19 measures on speech, language and communication abilities, social, and emotional functioning, mental health and changes in support services during the pandemic for CYP with Down syndrome specifically. Parents reported concerns about the negative impacts of COVID-19 restrictions across all domains. There were high levels of concern reported for learning, communication skills and social and emotional functioning and observations of deterioration in domains known to be areas of need associated with Down syndrome, such as expressive language and production of speech sounds/phonology and attention (Abbeduto et al., 2007; Grieco et al., 2015; Laws et al., 2015). Deterioration in wider domains related to social and emotional wellbeing and behavior, including greater reliance on adults and concerns about deterioration in social skills were also reported for some CYP with Down syndrome. This is consistent with other studies reporting data from parents/carers of children with SEND during the COVID-19 pandemic. In their sample of 241 parents of children with SEND, Asbury et al. (2021) reported parental worry about their children falling further behind in school as well as changes in behavior and low mood, in some cases severe challenging behavior. Broader impacts of COVID-19 have also been reported for adults with Down syndrome, including reduced independence and greater social withdrawal (Villani et al., 2020).

Relatively low levels of awareness of COVID-19 and worries related to COVID-19 were reported for CYP with Down syndrome. Moreover, there was a relatively small proportion of CYP with Down syndrome for whom parents/carers reported raised levels of anxiety since the onset of the pandemic (24%). This is in comparison with 73% in the remainder of the sample of CYP with SEND recruited as part of the larger study (Joffe et al., in preparation; Sideropoulos et al. 2022b) compared levels of anxiety for CYP with Down syndrome with CYP with other types of SEND, reporting lower levels of anxiety in CYP with Down syndrome, consistent with findings from this study. In line with these results, a population study (Van Gameren-Oosterom et al., 2011) has also reported lower levels of anxiety in children and young people with Down syndrome compared with other types of SEND and the general population. Increased rates of anxiety, however, have been reported in adults with Down syndrome (Vicari et al., 2013; Malegiannaki et al., 2019), and the younger age range of the sample in the present study may therefore also partly explain the lower levels of anxiety.

Anxiety was predicted by awareness of COVID-19 for both CYP with Down syndrome, and CYP with SEND (Sideropoulos et al., 2022a,b). Therefore, it is possible that the low awareness of COVID-19 reported by the parents/carers in our study could explain the lower rates of raised anxiety during the pandemic also reported. Moreover, low levels of awareness of COVID-19 may be in part explained by the difficulties parents/carers reported in explaining the pandemic and measures associated with COVID-19 to their CYP with Down syndrome. Asbury et al. (2021) report, that for children with a broad range of SEND, a lack of understanding and awareness of COVID-19 may have impacted on children with SEND differently, in some cases, resulting in lower levels of anxiety, and in others greater levels of anxiety, distress and challenging behavior due to a lack of understanding of the measures families were experiencing at that time. Parents/carers reported using a range of means to explain COVID-19 measures to their children, including social stories, telling stories and information provided by schools. Toseeb et al. (2020) similarly reported that some parents/carers of CYP with SEND felt that access to social stories to help explain the pandemic to their child with communication needs would have been helpful.

Most of the children with Down syndrome were not attending school during this period of the COVID-19 pandemic. Parents/carers reported concerns about meeting their child's needs and finding home-schooling difficult. Similarly, Sideropoulos et al. (2022a) reported higher levels of worry about school closures and change in routine for CYP with SEND compared with their typically developing siblings, and difficulties with home-schooling for CYP with SEND have been reported across a number of studies (O'Hagan and

Kingdom, 2020; Toseeb et al., 2020; Asbury et al., 2021). Asbury et al. (2021) found that parents of children with SEND reported feelings of distress and being overwhelmed by meeting their child's needs during school closures. Toseeb et al. (2020) suggest that this level of concern is likely to be much higher for parents of CYP with SEND due to their complex educational needs (although Toseeb et al. did not directly compare children with SEND with children without SEND). These findings have clear implications for additional education support needed by CYP and their parents/carers in the context of school closures.

High numbers of parents/carers reported a reduction, cessation or change in service delivery of specialist support services, including therapy services, psychology and specialist education services. This is consistent with reports of inadequate levels of support for children with SEND during COVID-19 (O'Hagan and Kingdom, 2020; Toseeb et al., 2020). Fifty percent of a sample of over 4000 parents/carers of CYP with SEND surveyed in May 2020 reported that external therapies had stopped (Disabled Children's Partnership, 2020). Community children and young people's services were particularly impacted by the COVID-19 pandemic at the time of this study, due to redeployment of staff to front-line services and changes in the mode of service delivery (Chadd et al., 2021; NHS Confederation and NHS Providers, 2022). This reduction in access to support services for CYP with Down syndrome, as for all children with SEND, arguably at a time of increased need, will have put them at increased risk of poorer outcomes, for example academic attainment, and difficulties with friendships, social functions and mental health (Royal College of Speech and Language Therapists, 2021). A deterioration in reading and writing skills, reported by 22% of parents/carers, suggests that education and speech and language therapy professionals may also need to pay particular attention to providing additional support to enable CYP with Down syndrome to reach their full potential in terms of literacy skills post-pandemic. Enhancing phonological awareness, decoding and providing positive home literacy environments have all been shown to be important and beneficial for promoting literacy in CYP with Down syndrome (Loveall and Barton-Hulsey, 2021).

Parents also reported that additional support for learning, speech and language, social–emotional and physical health would have been helpful during the pandemic, reflecting the complex pattern of needs and number of support services required for CYP with Down syndrome. This is similar to the finding of Toseeb et al. (2020), who found that parents/carers of CYP with SEND reported the need for ongoing support tailored to their child's needs as well as professional advice and support from teaching staff and therapies (Toseeb et al., 2020). It should be noted that in our study parents reported that social work and school counseling/pastoral care was much less likely to be stopped than other education and health services. It is interesting to consider why this was the case and how it was possible for some services to continue throughout the pandemic relatively unchanged. Learning and experiences from some services may be helpful in considering how other support services respond to a pandemic situation in the future.

It should also be noted that positive impacts of the COVID-19 measures were reported by some parents/carers of CYP with Down syndrome. These included increased time spent with siblings and family, improved play, communication and life skills. This is also consistent with findings from Asbury et al. (2021) who reported that, for some children with SEND, positive emotions were reported for children who found being home a respite from school, and O'Connell et al. (2020) who found that some parents reported improved quality time, increased family time and benefits to learning.

The present study measured changes in speech, language and communication abilities, behavior, social and emotional functioning, and mental health and access to education and healthcare services using a bespoke questionnaire and caregiver report rather than pre-existing validated measures. It is important that future work considers using validated caregiver measures to further explore associations between speech, language and communication abilities, and social emotional and behavioral outcomes in this population. This study focused on caregiver report of child outcomes rather than caregiver burden, stress and anxiety. An understanding of the impact of pandemic measures on caregivers, using measures such as the Caregiving Difficulty Scale (McCallion et al., 2005) would provide further insights. In addition the sample included parents of CYP with Down Syndrome across a wide age range. Due the relatively small sample size it was not possible to look at differences between age groups, but this would be important to explore in further studies with this population.

5. Conclusion

These findings highlight the wide-ranging impacts of COVID-19 on CYP with DS and their families, both in terms of impacts on learning, speech, language, communication and literacy as well as social, emotional and behavioral domains, and in terms of a lack of support from schools and community services. These findings have implications for the support that is now needed for CYP with Down syndrome and their families, who have experienced reduced provision throughout the pandemic, and important lessons for periods of social restrictions in the future. The high level of parental/carer concern about meeting their child's needs through home-schooling suggests the need for additional and individualized support for families of CYP with Down syndrome both during periods of school closures and also post-pandemic. Clear preferences to receive support from professionals, trained parents or a community of parents provide insights into new ways in which families can be supported, by widening the 'expert team' to include parents/carers and parent communities. Parents also seemed happy to receive support and training through a range of mediums, including online and virtual delivery and dissemination, making it possible for alternative solutions adopted as a result of the pandemic, and acceptable to families, to be incorporated into future mainstream provision of support.

Data availability statement

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

Ethics statement

Ethical approval was granted by the Faculty of Science and Health Ethics Sub-Committee at the University of Essex before the start of the study. The patients/participants provided their written informed consent to participate in this study.

Author contributions

EP, PH, and VJ contributed to the conception and design of the study. EP and RD organized the database. EP conducted the quantitative analysis. RD conducted and wrote up the content analysis. EP wrote the first draft of the manuscript. All authors contributed to manuscript revision, read, and approved the submitted version.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2023.1175636/full#supplementary-material

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Talkitt: toward a new instrument based on artificial intelligence for augmentative and alternative communication in children with down syndrome

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Introduction: Individuals with Down syndrome (DS) often exhibit a severe speech impairment, with important consequences on language intelligibility. For these cases, the use of Augmentative Alternative Communication instruments, that increase an individual's communication abilities, becomes crucial. Talkitt is a mobile application created by Voiceitt Company, exploiting speech recognition technology and artificial intelligence models to translate in real-time unintelligible sounds into clear words, allowing individuals with language production impairment to verbally communicate in real-time.

Methods: The study evaluated the usability and satisfaction related to the Talkitt application use, as well as effects on adapted behavior and communication, of participants with DS. A final number of 23 individuals with DS, aged 5.54 to 28.9 years, participated in this study and completed 6 months of training. The application was trained to consistently recognize at least 20 different unintelligible words (e.g., nouns and/or short phrases)/person.

Results: Results revealed good usability and high levels of satisfaction related to the application use. Moreover, we registered improvement in linguistic abilities, particularly naming.

Discussion: These results paves the road for a potential role of Talkitt application as a supportive and rehabilitative tool for DS.

KEYWORDS

language, machine learning, trisomy 21, digital application, speech

1. Introduction

Down syndrome (DS) is the most common genetic cause of intellectual impairment, estimated to occur once in approximately 1,000 births (Grimm et al., 2021). A high variability in the degree of cognitive impairment, ranging from profound to borderline intellectual functioning, is observed (Roizen, 2002; Vicari et al., 2013). Individuals with DS exhibit a neuropsychological profile characterized by weaknesses in the processing of verbal information associated with relatively spared visual information; moreover, they frequently exhibit delays in language development with receptive abilities usually more preserved than expressive abilities (Grieco et al., 2015). In particular, comprehension is usually related to the developmental stage, whereas expressive language quality is impaired in both vocabulary and syntax; for instance, it has been shown that syntax delays in individuals with DS are beyond expectations for cognitive level (Chapman and Hesketh, 2001; Eadie et al., 2002; Price et al., 2008). Speech production in DS is highly impaired: frequent sound errors with both protracted use of developmental phonological processes, such as final consonant deletion, and atypical phonological processes, presenting in association with inconsistent whole-word productions are frequently observed (Dodd and Thompson, 2001; Roberts et al., 2005; Cleland et al., 2010). Moreover, individuals with DS frequently omit words belonging to some grammatical categories such as prepositions or articles (Grieco et al., 2015). Altogether, these disorders compromise speech intelligibility. Over the years, a great number of risk factors have been identified for reduced speech intelligibility in DS, such as neuropsychological factors (e.g., short-term memory deficits), peculiar craniofacial features causing variations in laryngeal and resonator properties of speech, and hearing difficulties (Jarrold and Baddeley, 1997; Kent and Vorperian, 2013). However, there is no widely-accepted explanatory construct to sustain interventions for reduced intelligibility in DS (Faught and Conners, 2019; Wilson et al., 2019).

Overall, limited speech intelligibility represents a major issue in DS. Indeed, 95% of parents declared to be concerned about their child's ability to be understood (Kumin, 1994), and it has been documented that unintelligible speech in DS is a severer problem also in comparison with other intellectual disabilities (Rosin et al., 1988; Abbeduto and Murphy, 2004). Indeed, more than half of adolescents with DS have a hard time making themselves understood by anyone other than caregivers (Van Gameren-Oosterom et al., 2013). Moreover, important difficulties with morphosyntax and speech intelligibility can continue into adult age, hampering full participation in community life and independent living (Chapman and Hesketh, 2000). Therefore, supporting communication in DS represents a crucial aspect to promote socialization in such a population (Rodenbusch et al., 2013; Wilkinson and Finestack, 2020), supporting an amelioration in adaptive abilities and overall quality of life. Indeed, adaptive behavior includes conceptual, social, and practical skills required to function in everyday lives (Rapley, 2004). Communication skills, both comprehension and language production, are key component of social adaptive behaviors, allowing the individual to actively participate to the social environment he/she is included in.

Thus, since communication impairment plays a critical role in the development and social engagement of children with DS, it is fundamental to provide them with support aiding interaction processes. In particular, Augmentative and Alternative Communication (ACC) provides support to individuals with intellectual disabilities and complex communication needs (Beukelman and Light, 2020).

AAC is an assistive technology dedicated to patients with communication difficulties, which comprises several kinds of communication (other than verbal) and promotes all kinds of augmentative aids. According to the American Speech–Language–Hearing Association (American Speech-Language-Hearing Association, 2019), it is possible to classify AAC systems by distinguishing between aided and unaided systems. Unaided AAC systems involve the use of some parts of the body with communicative purposes, such as pointing, gestures, and facial expressions. Aided AAC systems involve the use of low-technology, such as symbol-based communication boards or books, or mid- and high-technology aids, such as speech-generating devices or electronic equipment (e.g., speech-generating devices, tablets with AAC applications; American Speech-Language-Hearing Association, 2019). AAC technology is evolving very rapidly. In the last years, AAC is taking advantage of a wide range of systems exploiting machine learning (ML) models to process and generate outputs by optimizing word prediction models and speech recognition algorithms; through ML, AAC systems can produce outputs in electronic digitized or synthesized speech (Elsahar et al., 2019). The relative affordability of mobile devices such as smartphones and tablets, associated with their portability and social acceptability (Still et al., 2014) makes high-technology AAC systems particularly suitable for individuals with developmental disabilities. Accordingly, some evidence on the preference of individuals with neurodevelopmental disorders for high-technology AAC has been reported (Ganz et al., 2013; Lorah et al., 2013; Couper et al., 2014). In their meta-analysis, Ganz et al. (2017) found low to moderate positive effects on social-communication outcomes for high-tech AAC use by individuals with intellectual and developmental disabilities throughout all school years. In particular, the meta-analysis reported that, despite some research demonstrating weak effects of AAC applications for this population, individual studies were significantly effective. The authors also considered a number of possible moderators to identify for whom and under what circumstances high-tech AAC implementation might be more or less effective. Results suggested AAC efficacy being independent from the implementer (i.e., researcher vs. natural communication partner), intervention context (i.e., natural vs. didactic contexts), behavioral strategies, age of participants, and communicative functions.

A recent systematic revision of the literature identified 12 AAC instruments for which some evidence of efficacy for people with ASD has been reported (Barbosa et al., 2018). In particular, systems such as Picture Exchange Communication System and Picture communication symbols seem to increase the interaction between individuals with DS and their peers, contributing to improving their quality of life and self-esteem (Barbosa et al., 2018). However, the authors highlighted the need for further well-designed studies investigating the effectiveness of various AAC devices to promote communication, socialization, and language abilities in DS. In particular, research investigating the effectiveness of high-technology AAC for DS is highly needed.

The present study was part of a broader project entitled "Speech recognition technology to enable people with Speech disabilities to communicate freely," funded by the Horizon 2020 program and coordinated by Voiceitt, a speech-recognition technology company. The Voiceitt team developed a customizable speech recognition system, the Talkitt application, a software application that translates unintelligible sounds into clear speech in real-time using a speech recognition algorithm. The purpose of the broader project was to optimize the Talkitt Basic application and validate it in multi-country trials on different populations. The optimization concerned the algorithm increased accuracy and recognition rates from 75% to 90%, overcoming error-free calibration and increasing discrimination, estimation of noise conditions and system stability. The validation in multi-country trials was aimed to demonstrate the applicability of the application (usability and satisfaction) in different environments and countries (Israel, United Kingdom, Spain, and Italy) with different languages, translating the user interface to ensure language independent use. The validation in different populations was aimed to demonstrate the applicability of the app (usability and satisfaction) and the impact on adapted behavior in people with different ages, diagnoses, and severities of speech disability (Acquired and developmental diseases, Traumatic brain injury, Stroke, Autism Spectrum Disorder, DS). Within the broader project, the present study aimed at training and demonstrating the applicability of the Talkitt Basic application in individuals with DS of Italian language. In particular, the present study aimed at training the artificial intelligence system dedicated to the recognition of vocal tracks spoken by individuals with SD with poorly intelligible language, and to the return of the correct interpretation of the audio track (audio reproduction of the word in real-time). The training of the system was necessary to build up a predictive mathematical model, based on ML, optimizing the speech recognition of 20 different unintelligible words/persons in our sample of children and adolescents with DS. To demonstrate the applicability, we evaluated the caregivers' satisfaction in using the Talkitt application and explored the impact of the use of the device on adaptive behavior. Communication abilities were also evaluated. The present study describes the results of the applicability of the application in our sample of individuals with DS, while the results on the algorithm improvement will be analyzed as part of the broader project results and will be described elsewhere.

2. Materials and methods

2.1. Participants

Participants with DS were recruited at the Child and Adolescent Neuropsychiatry Unit of a Children's Hospital in Rome. Italian was the primary language spoken at home for all participants. Inclusion criteria were as follows: diagnosis of DS confirmed by genetic testing; chronological age > 5 years; mental age ≥ 3.6 years; scores < 2 SD at the articulation subtest of Battery for Language Assessment in children aged 4 to 12 (Batteria di Valutazione Linguistica—BVL: 4–12; Marini et al., 2015). All participants were required to communicate verbally using a consistent language, but exhibiting moderate to severe phonological alterations; be comprehensible to closest relatives, at least in part, exhibiting consistent speech sounds; and having performed an otolaryngological examination that ruled out sensorineural hearing impairment and/or prescription hearing aids. Non-speaking individuals and youths with mild phonological alterations in expressive language (i.e., scores above −2 SD at the articulation subtest of Battery for Language Assessment) were excluded from the study. Participants' caregivers were also required to have an e-mail address to register with the Talkitt application and receive electronic communications while using it. Non-speaking individuals and youths with mild phonological alterations in expressive language were excluded from the study. We considered a language "consistent" when the same sequence of phonemes is produced each time it is uttered in the same context (e.g., picture naming). Phonetic variation in the production of a phoneme, captured by phonetic transcription, was not considered inconsistent. For example, [/a'mɛlla/] and [/a:a'mɛlla/], instead of [/kara'mɛlla/], *caramella* (candy), are two uncorrected forms but are not phonologically inconsistent, as the word is produced without a phonemic contrast each time (Bürki, 2018). Conversely, we considered a language as "inconsistent" when there are idiosyncratic words, words containing more than three phonological variations, the use of contrasting phonology, and the preference for one sound (e.g., theism). The assessment was made by an experienced clinical speech pathologist.

Thirty-four individuals with DS, aged 5.54 to 28.9 years (M = 11.25; SD = 5.19), participated in this study. Twenty-six participants were male and 8 were female. Intelligent Quotient (IQ) ranged between 43 and 69 (M = 56.37, SD = 8.13). Their diagnosis was molecularly confirmed by genetic test and all showed 21 free trisomy. All participants presented consistent language with different phonological and morphosyntactic difficulties and unintelligible speech. Out of the 34 participants, 23 (18 M, 5F) completed all the testing and the follow-ups, their average age was 9.44 years (ranging between 5.54 and 28.9 years; SD = 5.15) and their average IQ was 59.78 (ranging between 43 and 69; SD = 5.15).

2.2. Method

2.2.1. Talkitt application

Talkitt application is based on Voiceitt software that translates unintelligible sounds into clear speech in real-time using its proprietary speech recognition algorithm that recognizes unintelligible speech, and a large voice database of recordings from people with non-standard speech due to a variety of underlying conditions and disabilities.

The conceptual basis for the Voiceitt algorithms comes from the experiences of people with speech disabilities to date. It was observed that while people with these impairments struggle to be understood by outsiders, they are often understood with ease by family, friends, or caregivers who have learned how to adapt to their unique pattern of speech or prosody. From these observations, Voiceitt has been able to construct the Voiceitt algorithm to replicate this motion and recognize unintelligible speech. The innovation is in the recognition of unintelligible speech that requires powerful algorithms able to differentiate between indiscriminate sounds that are otherwise unintelligible to the human ear and standard speech systems.

Recognizing the complexity and the unique needs and characteristics of the users it serves, the core technology that forms Voiceitt's multi-layered solution is built upon main automatic speech recognition (ASR) architectures adapting state-of-the-art ASR techniques to recognize unintelligible speech:

Voiceitt Discrete ASR (DASR)—Voiceitt first-generation technology, called "discrete" speech recognition, is a customizable, language-independent, on-device ASR engine designed to suit highly unintelligible speech—i.e., what has been pre-calibrated by each user.

The Voiceitt iOS "discrete" application offers a personalized speech bank, or "dictionary." It requires the user to create and maintain a unique collection of their own specific words, phrases, and utterances. It is limited to the number of words or short phrases saved in the user's personalized dictionary, which they have chosen. Using the Voiceitt iOS application, the user will record words or phrases during the calibration phase to be stored in their personal speech bank and enable pattern matching based on prosody features. The more words added by a user, the greater level of recognition accuracy will be achieved. The application is limited to the communication content calibrated by the user, although she may pre-program an unlimited number of phrases into the dictionary. The user can also select the voice output, with the choice between adult-child and male–female voice outputs. Of note, the application does detect the differences in changes in the pronunciation of words or phrases. It tries to generalize the variations and map directly from a sound to a phrase/word and captures the speech variability intrinsically in the model. This requires the training set to be diverse so that it captures the typical variability (changes to pronunciations). The application also learns from the usage of the application. As a result, the examples that are used for training have increased variability over time. Therefore, during the on-boarding and training phase, participants actively "trained" the Artificial Intelligent system in recognizing selected words. Figure 1 shows how the algorithm identifies the patterns of sound unique to the unintelligible speech user.

Each individual user has a distinct phonetic inventory and adapting to their speech is similar to adapting the system for a new language. For each user, the larger collection of speech samples builds a better speech recognition model. The Talkitt algorithm is based on the estimation of speaker-dependent phonetic inventory by clustering similar sub-word linguistic units: upon launch of the application, an initialization requires the user to provide very small sample of recordings (five words, each repeated two times) which form clusters. The algorithm learns how the user says these specific words and recognizes them when spoken. In particular, the algorithm computes short-term signal energy of input each 20 ms over a window of 250 ms and, based on some pre-tuned threshold, it detects speech. If the computed energy is larger than a predefined threshold, start of a speech is declared; if it is the lower, end of speech is declared. As a unique user continues to use the application in this form, the algorithm steadily learns from the new recordings of the user that it receives.

By collecting a larger number of recordings from any one user, the algorithm can run clustering methods on the phonetic characteristics and identify units of sound in the user's unique speech style. This allows the mapping of these units in standard speech recognition and the application to a more extensive, unlimited vocabulary. Moreover, the user interface is intuitive, requiring minimum or null support for using it. See Figure 2 for user interface design.

2.2.2. Procedure

2.2.2.1. Enrollment, onboarding, and training

Enrollment. Participants were enrolled by requesting voluntary participation from families and children at the Child and Adolescent Neuropsychiatry Unit of a Children's Hospital and by involving DS associations operating at the national level. Informed consent was obtained from parents/caregivers of children and adolescents along with informed consent to the direct voluntary participant of the study.

Onboarding and training. For each participant, in agreement with caregivers, we selected 3–5 poorly intelligible words (nouns and/or short phrases) frequently pronounced by the child within specific daily life contexts (named scenarios). Families were invited to register about 20 repetitions/word for the entire duration of the project (6 months). Participants were instructed to pronounce the selected word naturally, without emphasizing specific parts of the word, thus avoiding, for instance, vocal emphasis, lengthening of syllables. Vocal data resulting from the registration were uploaded to the Voiceitt database to develop and improve the mathematical model of vocal recognition for each participant. Subsequently, parents could independently carry out onboarding for new additional words.

Through of the Voiceitt "Ambassador" software ("dashboard"), we carried out daily online remote monitoring to verify the use of the Talkitt application by the participants and, if necessary, to provide suggestions for the improvement of the voice model of each user. In particular, we checked the number of daily recordings, and the quality of the audio tracks and performed cutting of the essential track to the Artificial Intelligent System, when required.

2.2.2.2. Beta testing

Talkitt beta testing lasted for 6 months. It consisted of the daily use of the Talkitt device during the conversation. Participants were instructed to use the application daily (including the weekends) for 6 months, as much as possible within the scenarios chosen in the on-boarding and training phase. The mathematical model updated every time the patient pronounced the three words entered, recording the spoken vocal form (recording occurred only if the vocal form was recognized by the system). This training in the mathematical model made possible the continuous updating of the system of all the small modifications of the emitted vocal form. Researchers performed remote monitoring of the appropriateness of the vocal forms recorded by the system as corresponding to the words entered; they were able to accept or reject the inclusion of new recordings in the mathematical model of speech recognition of the initial three words. After obtaining

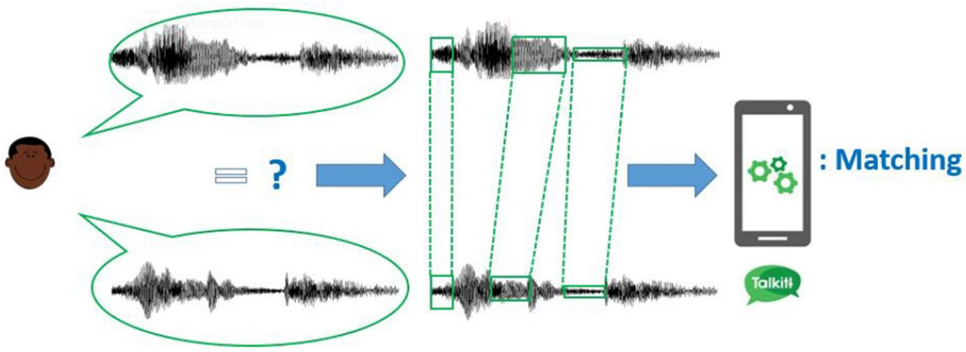
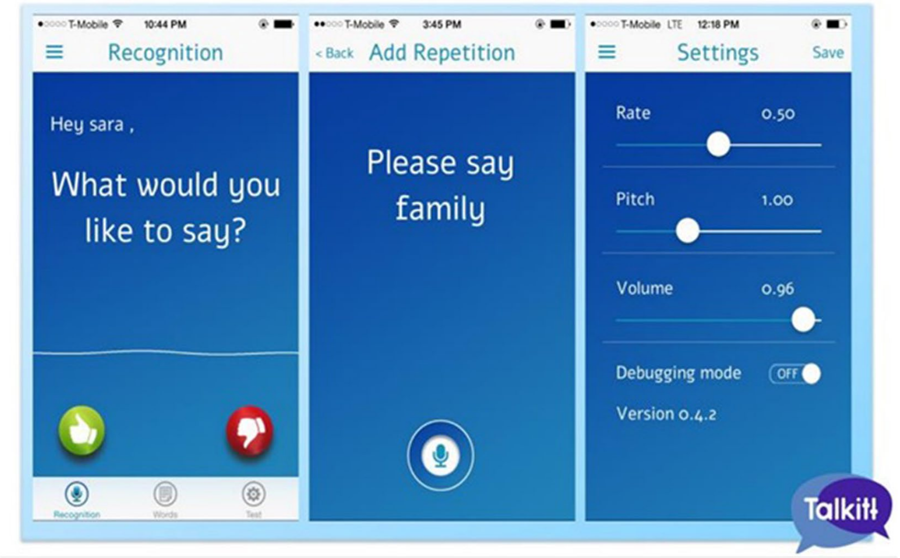


FIGURE 1
Illustration of how the Talkitt algorithm identifies the patterns of sound unique to the unintelligible speech user and use frame matching to map those consistencies to standard speech and the recorded sounds, thus successfully translating unintelligible speech. This figure is reproduced with permission from the Voicett company.



Hey sara ,

What would you like to say?

Please say family

Rate 0.50

Pitch 1.00

Volume 0.96

Debugging mode OFF

Version 0.4.2

FIGURE 2
User interface design. The application is intuitive, requiring minimum or null support for usage. This figure is reproduced with permission from the Voicett company.

a sufficient number of repetitions for each word (about 20), vocal forms of other words were recorded, up to about 20 words. The procedures for inserting the vocal forms into the mathematical model follow the abovementioned procedure. To have a variety of vocal samples and diversified confusing background noises, participants and caregivers were instructed to use the Talkitt device at various times of the day. During the 6 months, researchers were constantly in touch with engineers of the Voiceitt team to face possible issues. In the automatic phase, i.e., when the Artificial Intelligent System has now learned the unintelligible words of the child or teenager, the Talkitt application has reproduced the correct spoken vocal form. At the same time, to refine the training of the system, through the same Talkitt application, the child, the parent, or the child's therapist could confirm whether the interpretation of the system was correct or not.

2.2.2.3. Re-test

The intermediate re-test consisted of an application setting check to verify the technical functionality. The final re-test consisted of the administration of standardized tests administered during the enrollment phase 6 months after the start of the use of the Talkitt application.

2.2.2.4. Follow-up

The primary outcome was the applicability and was assessed by means of a Satisfaction questionnaire at the end of the trial, i.e., 6 months after the onboarding phase. An intermediate measurement was carried out at the end of 1 month from the onboarding phase, to monitor the level of accuracy of recognition of the model and propose any technical adjustments. These results were not analyzed in the present study. The secondary outcome was the impact of the application use and was measured by the Adaptive Behavior questionnaire during the onboarding phase and at the end of the experimentation, after 6 months. Language abilities was also assessed at follow-up.

2.3. Measures

2.3.1. Nonverbal intelligence

The Leiter-3 (Leiter et al., 2013) offers a nonverbal measure of intelligence and evaluates the ability to reason by analogy, matching and perceptual reasoning in general, irrespective of language, and formal schooling. This makes Leiter-3 particularly suitable for individuals with language difficulties. The nonverbal IQ obtained from the Leiter-3 is based on four subtests: Figure Ground, Form Completion, Classification and Analogies, and Sequential Order.

2.3.2. Applicability: satisfaction questionnaire

The satisfaction questionnaire for the Talkitt device, filled by the user or the caregiver, consisted of 18 multiple-choice questions about the usability and frequency of use of the device. The following areas were investigated: quality of the instructions received for the use of the device, easiness and frequency of use, pleasantness of the interface, contents' clearness, quantity of added words, the improvement of language production, and overall satisfaction were investigated.

2.3.3. Language assessment

The Battery for Language Assessment in children aged 4 to 12 (Batteria di Valutazione Linguistica—BVL_4–12; Marini et al., 2015) systematically assesses phonological, lexical, semantic, pragmatic, and discursive skills in production, comprehension, and oral repetition tasks in children and adolescents, detecting communication and linguistic disturbances. This linguistic assessment scale comprises three sections: the assessment scale of oral production skills, the assessment scale of oral comprehension skills, and the oral repetition scale. To evaluate the articulation abilities of the participants and the intelligibility level of the speech, we used BVL_4–12, naming, and articulation subtests. Cronbach's alpha are good for all age groups: mean values for naming range from 0.80 to 0.81, for articulation is 0.87.

2.3.4. Adaptive behavior

The ABAS-II parent-report measure (Oakland, 2008) was used to assess the individual's daily adaptive functioning. Parents or caregivers were asked to assess how often their child engages in a particular activity using a 4-item Likert scale (0—is not able, 1—never when needed, 2—sometimes when needed, and 3—always when needed). The measure consists of 10 skill areas: communication, community use, functional academics, home living, health and safety, leisure, self-care, self-direction, social, and work skills. ABAS-II provides norm-referenced standard scores for three domains: conceptual domain (CON), social domain (SOC), and practical domain (PRA) and a merged score—general adaptive composite (GAC)—(M 100, SD 15, and 90% and 95% confidence intervals and percentile ranks). Reliability coefficients for the general adaptive composite are in the high 0.90s for all age groups, ranging from 0.97 to 0.99. Reliability coefficients for the adaptive domains range from 0.91 to 0.98. Average reliability coefficients of the skill areas across age groups range from 0.85 to 0.97. Here, we provide some examples of the items included in the questionnaire: "Speaks clearly" (Conceptual domain), "carries scissors safely" (Practical domain"), and "says please when asking for something" (Social domain).

2.4. Statistical analyses

Descriptive statistics were used to describe the demographic characteristics of the participants. To evaluate the effect of Talkitt beta testing, repeated measure analysis of variance—ANOVA—has been performed on the ABAS II subscales composite scores and the BVL 4–12 tests raw scores, between T0 (before testing) and T1 (after 6 months of testing). The sphericity assumption, verified by Mauchly's sphericity test, has been met. Bonferroni correction for multiple comparisons was applied so that the significant difference was set at the $p<0.0042$ level. Partial eta squared (ηp2) was used to measure effect size. Outliers on the improvement, i.e., the difference between T1 and T0, were evaluated per each variable to identify if any participant benefited to a greater extent from the application. The Z-value was calculated, considering the mean and the standard deviation of the difference. The data with Z-values beyond 3 were considered as outliers. We identified only one outlier value: in particular, one participant showed a greater improvement in the ABAS Adaptive behavior Practical domain (Z-value = 3.9). To avoid any bias, this observation was dropped from the analyses.

2.5. Ethical considerations and data storage

The protocol was in full compliance with the Helsinki Declaration, and it was approved by the local Ethical Committee (163_OPBG_2018). Voiceitt manages a voice database of people with speech problems ("Impaired Speech Corpus"), an essential component to improve research and development of more advanced methods for recognition of speech impairment. Recordings of Talkitt users were continually copied to the database and the processes noted in the Library. No personally identifiable information has been transferred to this database. According to the new 2016/67 European Regulation, which entered into force on 25 May 2018, personal data, i.e., the e-mail address and voice recording, useful for using the Talkitt application or for monitoring technical problems, has been processed in the more absolute respect for the principles of correctness, lawfulness, relevance and non-surplus envisaged by art. Eleven of the aforementioned legislative decree, using IT tools, adopting suitable measures to guarantee the security and confidentiality of the data and will be kept for the time necessary and instrumental for the pursuit of the project's purposes. Participants' data are available only for the Data Processor and his collaborators. The participants' personal data will not be disseminated.

3. Results

The accuracy of the application ranged between 60% to 95% according to the participant's impairment. However, the results on the algorithm accuracy improvement will be analyzed as part of the broader project results and will be described elsewhere.

3.1. Application usability

Application usability and functionality were assessed through a questionnaire collected from the parents. Data of the mean percentage of response are reported below (Figure 3).

As is shown, more than 70% of participants reported the highest level of satisfaction with: "Clear instructions," "Easy registration and login," and "Font size." More than 70% of participants reported a medium satisfaction level for: "Easy word registration," "Easy automatic phase," "Used weekly," "Daily usability," and "Improves linguistic production." Finally, more than 70% of participants reported the minimum level of satisfaction for: "Use at school."

Of note, the management of the application was intuitive enough to require minimum or null support from caregivers.

3.2. Effect of Talkitt use on linguistic and adaptive functioning

Adaptive behavior was assessed by the ABAS II subscales composite scores after 6 months of Talkitt use. Although a general improvement was observed in all subscales, significant amelioration emerged in the Global composite score with a medium effect size. Similarly, a significant amelioration emerged in the verbal abilities, evaluated through the BVL 4–12 tests. Although both Naming and Articulation raw scores improved, only Naming improvement survived after Bonferroni correction and with a medium effect size (see Table 1 for details).

4. Discussion

This is the first study to report the satisfaction and the effects on linguistic abilities of an ML-based CAA intervention in children with DS and their caregivers who have participated in a 6-month beta test. The purpose of this work was to use Talkitt Application to train an Artificial Intelligence System in recognizing 20 unintelligible words/person in a sample of youths with DS. We also evaluated the caregivers' satisfaction with using the Talkitt application and device. Finally,

FIGURE 3
Parents report satisfaction with Talkitt use.

TABLE 1 Adaptive and linguistic measures at follow-up.

Test	Subscale	T0 mean	(SD)	T1 mean	(SD)	$F(1,22)$	p	η^2
ABAS adaptive behavior	Global[a]	55.6	14.2	62.4	17.0	10.78	0.003*	0.33
	Conceptual[a]	57.7	11.4	56.9	12.4	0.10	0.76	0.004
	Social[a]	71.4	15.8	76.4	17.9	4.18	0.05	0.16
	Practical[a,b]	56.2	17	60.5	19.7	8.75	0.008	0.29
BVL language production	Naming[c]	36.6	16.8	45.1	15.5	20.46	<0.001*	0.48
	Articulation[c]	30.1	29.3	40.0	35.2	4.71	0.04	0.18

[a]Standard score; [b]$F(1,21)$; [c]row score.
*Significant after Bonferroni correction.

we investigated the possible benefits of adaptive behavior and language abilities deriving from the use of Talkitt application/device. The algorithm was trained correctly. Overall, caregivers of children who completed 6 months of the beta test were satisfied with the Talkitt application/device as a means to improve communication abilities. Caregivers perceived Talkitt as easy to use and beneficial to their children. The evaluation of language skills in our sample confirmed such perception, demonstrating an effective improvement of oral production in our sample. We also detected an amelioration of global adaptive abilities.

4.1. Talkitt has a good usability and exhibited high levels of satisfaction among caregivers

A majority of caregivers reported being very satisfied or satisfied with the following items: clearness of the provided instructions for the use of the device; the ease of registration and login; the pleasantness of the graphical interface; clearness of the provided contents; the ease in adding new words to train the algorithm; the easiness in registration new words; intuitiveness of the device/application; the sensitivity of the automatic recognition; the font size; the added words; the weekly use; the improvement of linguistic abilities; and overall satisfaction. Of note, the management of the application was intuitive enough to require minimum or null support from caregivers. Taken together, these results depict high levels of Talkitt usability and perceived usefulness. The delivery of an ML-based CAA intervention in children with DS is an innovative yet strategic approach, as it allows for overcoming barriers to interaction specific to this population. Automated speech analysis is a useful tool for analyzing and modifying speech in speech disorders, also in pediatric age (McKechnie et al., 2018). Children who need speech therapy could have significant barriers, given that this kind of intervention is often costly and time-requiring (McAllister et al., 2011) and caregivers could need alternative systems to gain access to services (Ruggero et al., 2012). Technology-based approaches can be elective tools to overcome these issues since they allow temporal and local independence, easy accessibility, and scalability (Ebert et al., 2018). Moreover, interventions based on the ML approach provide tailored support, helping to define the most appropriate course of action for a patient.

In particular, since Talkitt is an intelligent AAC solution, it may easily predict the language abilities of children with DS even if the input could be in part erroneous and incomplete. This aspect is of great advantage in comparison with the conventional classification methods (Thomas et al., 2017) because it can increase the probabilities of incoming words and phrases, and complete sentence transformations (Higginbotham et al., 2011) leading to a more proficient conversation.

Another advantage of intelligent AAC systems consists in the improvement and ease of use of devices and the associated user interfaces (Elsahar et al., 2019). The focus on the user activity to be carried out needs to be at the core of the implementation. Talkitt application shows high usability since its use does not require voluntary muscle controls, but the device is activated by the simple vocal recognition of the target words. This easiness of use could be of crucial relevance in cases of ID and possible difficulties in instruction understanding or executive functions (Costanzo et al., 2013).

Finally, affordability, in terms of costs associated with the hardware and software requirements of the utilized device, and portability, in terms of easiness of moving, have a great impact on the AAC device use (Elsahar et al., 2019). Since Talkitt is an available application, easy to download for different kinds of common-use devices such as smartphones or mini-tablets, it is suitable for usage in different settings.

However, the item concerning the use of Talkitt at school obtained the lowest score. In particular, caregivers referred to difficulties in verbal exchanges with classmates. Since the application was trained for a low number of words (about 20), which did not cover the entire vocabulary, it is possible that this could have limited the interactions in an unfamiliar environment.

Recently, the Voiceitt technology has evolved and uses a technology capable of updating itself more easily and of traslating entire sentences. Voiceitt's next-generation technology recognizes "continuous" speech, i.e., vocabulary that has not been pre-calibrated. Surely, the use of an advanced level of the application could allow greater integration in environments outside the family and therapeutic one and could represent a valuable future development of the Talkitt application in DS.

4.2. Talkitt application/device improved adaptive abilities and language in children with DS

Talkitt application/device improved adaptive abilities in children with DS. Our results show a significant effect of the use of the Talkitt application/device on adaptive abilities, evaluated through ABAS II. Adaptive skills are defined as "the effectiveness with which the individual copes with the natural and social demands of his environment" (Heber, 1959). Thus, adaptive behavior supports autonomous functioning across several daily contexts and responsibilities (Tassé et al., 2016). In addition to impairment in cognitive and language abilities, children with DS exhibit important limitations in adaptive behavior. The limitation in adaptive behavior could be a direct consequence of reduced language abilities. Indeed, from an early age, children with developmental disabilities who have limited speech are strongly limited in participating in language and literacy instruction and social interaction; moreover, they are known to be at a greater risk for limited development of these skills for reasons both intrinsic to their disability and extrinsic to their learning environment (Ogletree, 2021). A recent review of the literature therefore highlights how AAC intervention can support not only vocabulary development and expressive language, but also social communication and adaptive behavior since preschool age (Allen et al., 2017; Ogletree, 2021). A possible explanation why general adaptive functioning may have improved after the application use could be related to a potential increase on participation, an essential dimension of human functioning according to the American Association of Intellectual Disabilities and Developmental Disabilities (Buntinx and Schalock, 2010). Participation includes social roles, involvement in leisure activities, choice, and control. A facilitation in communication exchanges by the application, may have indirectly affected the degree of participation and in turn the general adaptive functioning.

Although unexpected, we also found an improvement in linguistic abilities, in terms of speech and vocabulary improvement. The use of several different AAC systems (Binger et al., 2010; Kent-Walsh et al., 2010; Quinn et al., 2020) has been demonstrated to increase expressive vocabulary in children with DS. One possible reason could be that using AAC devices during a conversation would prompt the child to answer and elicit a response with the target word based on the AAC device, and that these strategies could support their vocabulary development (Ogletree, 2021). Given the positive outcomes that AAC application has had for vocabulary, language, and social communication development for children with DS, the use of AAC from a very early age seems promising. Moreover, given the high portability of the Talkitt device, i.e., the easiness of moving the device for usage in a different setting, its usability could be high for different contexts and different ages.

4.3. Limitations

However, this work is not without limitations. First, we evaluated only linguistic abilities at follow-up and no information is available on other cognitive measures. We cannot therefore exclude that the unexpected effect on naming improvement could be related to a general development or to other cognitive abilities changes. Further, it cannot be ruled out that the observed positive effects in naming abilities could have also positive effects on other cognitive domains; future studies are required to investigate these hypotheses. Finally, another limitation of the study is the lack of information on applicability (usability and satisfaction) and adaptive behavior impact from the participants themselves. This information is very important for understanding the individuals' views and will need to be collected in future testing of new versions of the application.

Moreover, the use of the application did not exclude other concomitant treatments and was provided in addition to as usual activities. Another limitation of the study was not controlling for the type and amount of therapeutic and extracurricular activities of each participant. These aspects may have interacted with the effects of the app. It cannot be ruled out that the use of the application complemented with speech therapy could produce additional benefit on communication skills of children with DS. Future studies should take into account possible synergistic effects of the use of Talkitt plus concomitant treatment and activities.

Finally, there is a limitation of the present version of the application: the speech recognition is designed to recognize discrete words or phrases, for which the participant/user has provided adaptation data (or "seen data") belonging to an individualized library. This limitation could lower spontaneity and fluency of the speech. To overcome this limitation, the algorithm should have the capability to recognize phrases for which the user has provided no data, or "unseen phrases." Recent algorithmic developments of the app (Voiceitt next-generation technology) has evolved from discrete to continuous speech recognition, recognizing "continuous" speech, i.e., vocabulary that has not been pre-calibrated. This could lead to greater spontaneity and fluency without being limited to a closed vocabulary of pre-recorded phrases. This advanced version should be tested in future study to prove the accuracy and usability in population with DS.

4.4. Conclusion

These positive results and the high compliance emphasize the feasibility and efficacy of an ML-based AAC intervention for the improvement of communication abilities and adaptive abilities promotion for children with DS. Moreover, the advances in the integration of AAC systems with Artificial Intelligent applications could improve access to high-tech devices, the speed of output generation, and the customization and adaptability of the AAC interfaces to suit the needs and requirements of each individual user. Finally, the use of the present application could help expand the scope of AAC beyond physical communications, increasing the usability and the context of usage of future AAC solutions for children with DS.

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 Bambino Gesù Children's Hospital Ethical Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

FC, SV, SS, and DW: conceptualization. FC, CC, SS, and DW: methodology. FC and CC: formal analysis. CC, DR, and FC: investigation. CC and DR: data curation. EF, FC, and CC: writing—original draft preparation. EF, FC, CC, SS, DW, and SV: writing—review and editing. SV: supervision and project administration. All authors contributed to the article and approved the submitted version.

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Conflict of interest

DW and SS have a commercial and financial interest in the Talkitt application, since they are, respectively, CoFounder and CEO and Director of Business Development of Voiceitt.

The remaining authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Parent-implemented augmented communication intervention and young children with Down syndrome: an exploratory report

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Introduction: Young children with Down syndrome (DS) present with speech and language impairments very early in childhood. Historically, early language intervention for children with DS included manual signs, though recently there has been an interest in the use of speech-generating devices (SGDs). This paper examines the language and communication performance of young children with DS who participated in parent-implemented communication interventions that included SGDs. Specifically, we compared the functional vocabulary usage and communication interaction skills of children with DS who received augmented communication interventions (AC) that included an SGD with those children with DS who received spoken communication intervention (SC).

Methods: Twenty-nine children with DS participated in this secondary data analysis. These children were part of one of two longitudinal RCT studies investigating the effectiveness of parent-implemented augmented communication interventions in a larger sample of 109 children with severe communication and language impairments.

Results: There were significant differences between children with DS in the AC and SC groups in terms of the number and proportion of functional vocabulary targets used and the total vocabulary targets provided during the intervention at sessions 18 (lab)and 24 (home).

Discussion: Overall, the AC interventions provided the children with a way to communicate via an SGD with visual-graphic symbols and speech output, while the children in the SC intervention were focused on producing spoken words. The AC interventions did not hinder the children's spoken vocabulary development. Augmented communication intervention can facilitate the communication abilities of young children with DS as they are emerging spoken communicators.

KEYWORDS

Down syndrome, language development, speech development, augmentative and alternative communication (AAC), early intervention

Introduction

Children with Down syndrome (DS) are known to present with speech and language impairments (Abbeduto et al., 2007; Romski et al., 2021; Wilkinson and Feinstack, 2021). These speech and language impairments are evident very early in childhood with the delayed onset of babbling followed by a gap between receptive and expressive language development

(Miller, 1999; Fidler, 2005; Romski et al., 2021; Wilkinson and Feinstack, 2021). The most striking finding is that children with DS often have relatively strong receptive language skills but have more significant delays in expressive language skills (Miller, 1999; Warren et al., 2020).

One intervention approach that has been part of early language interventions for young children with DS is the use of augmentative and alternative communication (AAC; Wilkinson and Feinstack, 2021). The American Speech Language Hearing Association (ASHA, 2019) defined AAC as an area of clinical practice that addresses the needs of individuals with significant and complex communication disorders characterized by impairments in speech-language production and/or comprehension, including spoken and written modes of communication. AAC includes unaided and aided forms of communication. Unaided forms of AAC include simple gestures, facial expressions, and other manual signs. Aided forms of AAC include picture communication boards, dedicated computers that talk using a synthetic or digitized voice (also described as speech-generating devices or SGDs), as well as iPads or other tablets with various software applications (Beukelman and Light, 2020).

Historically, the use of manual signs, or unaided AAC, was employed with young children with DS who demonstrated emerging spoken language skills (Bird et al., 2000). Manual signs were thought to serve as a bridge to early receptive and expressive spoken language (Iverson et al, 2008). Studies supported this rationale and found that young children with DS could learn to use manual signs to communicate (Romski and Ruder, 1984; Kouri, 1989; Launonen et al., 1996; Foreman and Crews, 1998; Wright et al., 2013; Kaiser and Hampton, 2017). Key word signing has been used successfully with older children with DS (Frizelle and Lyons, 2022). Parents, however, reported a range of issues that may impact the success of key word signing (Glacken et al., 2019). There are two important clinical issues related to manual sign instruction (Romski et al., 2021). First, children with DS have difficulty continually producing intelligible signs given their motor dexterity difficulties. Second, communicative partners of children with DS must learn to understand and produce manual signs, thus potentially limiting the number of communication partners who can understand and use manual signs with this population.

Recent technological advances in aided forms of communication may provide a choice that complements unaided forms of AAC. These aided forms of communication place different motoric demands on the child with DS by having the child point to or touch a symbol on a board or device rather than physically produce a manual sign. When speech output is available, the partners also hear the spoken word, albeit synthetic or digital. In a case study, Iacono and Duncum (1995) found that the use of speech output AAC technology paired with manual signs was more effective than manual signs alone for eliciting single-word productions as well as two- and three-word combinations. In recent years, SGDs have been increasingly used with children with developmental disabilities to support communication development during early intervention and preschool (Romski et al., 2015; Barton-Hulsey et al., 2021). The outcomes of this work suggest positive gains in receptive and expressive communication for children with developmental disabilities. Less attention has been focused specifically on children with Down syndrome and the outcomes associated with the use of SGDs compared to other communication intervention approaches (Barbosa et al., 2018). There are a few studies that focus on the use of SGDs with children with developmental disabilities (Van der Meer et al., 2012; Barbosa et al., 2018). In two randomized controlled trials, Romski et al. (2010, 2023) found that augmented communication interventions that included an SGD and parent coaching had a positive effect on communication for young children with developmental delays who began intervention with less than 10 spoken words. The positive effects included increases in vocabulary size, the spontaneous use of targeted symbol vocabulary, and communication interaction skills. Importantly the augmented communication interventions did not hamper spoken vocabulary development.

While some of these studies found that SGDs are viable for use with children with developmental disabilities, they did not focus exclusively on use with children with DS in the service of language intervention. Additional research is needed to examine the role SGDs can play in early language interventions for young children with DS.

This paper explores the language and communication performance of young children with DS who participated in a larger randomized controlled study of parent-implemented communication interventions that included SGDs. Specifically, compared the functional vocabulary usage and communication interaction skills of children with DS who received augmented communication intervention with those who received spoken communication intervention alone. This study also compared parent communication interaction skills between intervention groups after participating in coaching in the lab and at home.

Method

Participants

Twenty-nine children diagnosed with DS were included in a secondary data analysis. These children already participated in one of two longitudinal randomized controlled studies of parent-implemented communication interventions as part of a larger sample of 109 children with severe developmental delays and communication and language impairment that did not separately report on the performance of the 27% of children with DS. Children were recruited from a variety of professional sources in the metropolitan Atlanta area who provided services for children with communication and language impairment, including clinical psychologists, developmental pediatricians, pediatric neurologists, and SLPs. The subset of children with DS included 20 males and 9 females, ages 24 to 29 months ($M = 28.67$, $SD = 3.65$) at the onset of the study. All children had an expressive language age-equivalent score on the *Mullen Scales of Early Learning* (MSEL; Mullen, 1995) of less than 12 months and a vocabulary of no more than 10 spoken words according to parent reports and clinical observations. Other inclusion criteria were a primary language of English and the presence of sufficient gross motor skills to manipulate an SGD.

A set of developmental assessments, including measures of communication, adaptive behavior, and motor, and visual–spatial reception skills was administered to each child during pre-intervention by a certified SLP who was masked to the child's group assignment. The baseline developmental assessment provided a description of the children's developmental and language skills at the onset of the study. Pre-intervention assessment scores for all children with DS were reported by augmented or spoken communication intervention groups in Table 1. Standard scores on the MSEL visual reception and

TABLE 1 Pre-intervention assessment scores for children by intervention group.

	Intervention group	
Assessment variable	**SC (n =6)**	**AC (n =23)**
Age (months)	28.17 (1.72)	28.78 (4.02)
VABS ABC (SS)	64.17 (6.15)	70.30 (6.47)
MSEL ELC (SS)	51.67 (4.76)	57.78 (9.97)
MSEL visual reception (SS)	21.83 (3.0)	28.74 (10.78)
MSEL fine motor (SS)	24.0 (6.48)	22.74 (5.88)
MSEL receptive language (SS)	21.0 (2.53)	28.13 (9.33)
MSEL expressive language (SS)	22.33 (3.83)	23.70 (5.35)
SICD receptive language (months)	16.67 (3.93)	19.18 (4.73)
SICD expressive language (months)	15.67 (2.66)	18.82 (5.68)
MCDI spoken words	11.83 (10.61)	15.70 (17.70)
MCDI words understood	178.17 (145.05)	132.87 (70.13)

SS, standard score; SC, spoken communication group; months, age equivalent score in months; AC, augmented communication group; VABS, Vineland adaptive behavior scales; MSEL, Mullen scales of early learning; SICD, sequenced inventory of communication development; MCDI, MacArthur-Bates communicative development inventories; Scores are means (with standard deviations in parentheses).

receptive language domains were significantly higher for children in the augmented communication group than the spoken communication group, $p = 0.012$ and $p = 0.003$, respectively. There were no significant differences in expressive communication skills and no other significant group differences on any other baseline measures.

At the time of participation in each of the original studies, 28 of the 29 children with DS (97%) were receiving speech-language therapy for an average of 1.1 (SD = 0.6) hours per week, and 17 children with DS (59%) were reported to be using at least a few manual signs to communicate. It is important to note that the intervention provided in these two studies was supplemental to the clinical services that were received outside of participation in the study.

Overview of randomized-controlled studies

In the first study, 62 parent–child pairs, including 18 children with DS (29% of the sample), were randomly assigned to a spoken communication intervention or one of two augmented communication interventions that employed the use of an SGD (Romski et al., 2010). In the second study, 47 parent–child pairs, including 11 children with DS (23% of the sample), were randomly assigned to one of two augmented communication interventions (Romski et al., 2023).

Description of interventions

The overarching goal of all the interventions was to increase the spontaneous use of the target vocabulary words and to engender improved support for communication interaction between the child and parent at home. In both of the two original studies, parent–child pairs were assigned, via a randomized stratification strategy for etiology and MSEL composite score, to one of four intervention groups: Spoken Communication (SC, DS $n = 6$), Augmented Communication- Input (AC-I, DS $n = 6$), Augmented Communication- Output (AC-O, DS $n = 12$), and Augmented Communication-Input Output Hybrid (AC-IO; DS $n = 5$). As seen in Table 2, all four interventions shared a common structure and vocabulary and the three augmented interventions shared the same mode (using an SGD to include visual-graphic symbols with speech output). The SC intervention focused on developing spoken language vocabulary words. In order to maximize the sample size in the current study, the three augmented communication groups were combined and referred to as the Augmented Communication (AC) group.

Each intervention was designed to be completed over the course of 12 weeks (two sessions per week), with the first 18 sessions (9 weeks) occurring in the Toddler Language Intervention Project Lab at Georgia State University, and the final 6 sessions (3 weeks) occurring in the children's homes. Each 30-min intervention session consisted of natural communication interactions during three 10-min activities. The three child-oriented activities were (1) playing with toys, (2) reading/looking at picture books, and (3) eating a snack in that order. These three activities were designed to simulate routines that the parent and child engaged in at home.

As detailed in Romski et al. (2010, 2023), at the beginning of each of the 12 weeks parents received training materials that detailed the parent and child goals for that week. Parents were gradually guided through the activities by the SLP as they observed the session conducted by one of nine trained interventionists (with a bachelor's degree or higher in communication or psychology) through a one-way viewing window. As the parent backed into the session's activities beginning with the snack, the interventionist coached them to conduct the activities until they were leading the entire intervention session. See Table 3 for an overview of parental participation across all sessions. The guidance began with parent observation and gradually brought the parents into the intervention sessions. Beginning with parent observation was based on parent feedback during a pilot intervention implementation. As part of using the SGD or spoken intervention strategy, the interventionist and SLP coached the parents to integrate naturalistic communication intervention strategies during the three activities. These strategies created communication opportunities for the child to use target vocabulary during the activities (e.g., offering choices, pause time, and environmental arrangement of toys, books, and snacks to create communication temptations). Parents received guidance and coaching when needed from the interventionist and the SLP throughout the course of the intervention sessions. At the end of each session, the parent, interventionist, and SLP discussed the outcome of the session.

Target vocabulary word selection

Each parent worked with the SLP to select a set of target vocabulary words (words the child did not comprehend or produce in speech or manual sign), motivating and appropriate to the three activities, that would appear on the SGD or be spoken during the sessions. None of the target vocabularies included manual signs. If a child had a manual sign for a vocabulary item, it was not included as one of their target vocabulary items.

At the onset of the intervention, each child had approximately 16 target words. Examples of these target words included during play: *doll, car, push*; picture book reading/looking: *open, book, dog*; and snack: *cookie, juice, more*. Some of the target vocabulary words were

TABLE 2 Description of intervention by group.

	Intervention group			
Intervention component	SC	AC-I	AC-O	AC-I/O
Target vocabulary	I/P and child use speech to communicate	I/P uses the SGD to provide comm. Input to child	Child uses the SGD to communicate	I/P uses the SGD to provide comm. Input; the child uses SGD to communicate
Mode	Individualized vocabulary of spoken words	Individualized vocabulary of visual-graphic symbols + words	Individualized vocabulary of visual-graphic symbols + words	Individualized vocabulary of visual-graphic symbols + words
Strategies	I/P encourages and prompts the child to produce spoken words	I/P provides vocabulary models to child using the SGD; Symbols are positioned in the environment to mark referents	I/P encourages and prompts the child to produce communication using the SGD	I/P provides vocabulary models to child by using the device; Symbols are positioned in the environment to mark referents; I/P encourages and prompts the child to produce communication using the SGD
Parent coaching	I provides resource and coaching for P	I provides resource and coaching for P	I provides resource and coaching for P	I provides resource and coaching for P

SC, spoken communication; AC-I, augmented communication-input; AC-O, augmented communication-output; AC-I/O, augmented communication-input/ output; I, interventionist; P, parent. Adapted from Sevcik et al. (2021).

specific to one of the three activities (e.g., doll) and others were used across all three activities (e.g., more). When the parent and SLP determined that the child was consistently using a target vocabulary word appropriately during the intervention sessions, new words were added either to the SGD or to the spoken vocabulary list.

Measures

A number of measures were obtained from the children and their parents over the course of the study. First, a set of developmental assessments were administered to each child at the pre-intervention sessions. Second, intervention outcomes were measured by assessing growth in spontaneous target vocabulary production and communication use at the end of the intervention (sessions 18 and 24). Manual signs were not included in any of the measures.

Developmental assessments battery

The set of developmental assessments administered included measures that were directly administered to the child as well as measures that solicited parent reports of communication skills. These measures provided a profile of the children's developmental skills. The following measures were administered: *The Mullen Scales of Early Learning* (MSEL; Mullen, 1995) is a clinician-administered developmental measure that assesses a child's gross motor, fine motor, visual reception, and expressive and receptive language skills. The *Vineland Adaptive Behavior Scales* (VABS; Sparrow et al., 1994; VABS-II; Sparrow et al., 2005) are parent interviews that measure adaptive functioning across four domains: communication, daily living skills, socialization, and motor skills. The *Sequenced Inventory of Communication Development* (SICD; Hendrick et al., 1984) was also administered as an additional measure of expressive and receptive communication that includes direct assessment supplemented by parent reports. The *MacArthur-Bates Communicative Development Inventory: Words and Gestures* (MCDI; Fenson et al., 1993) was administered as an additional measure of parent-reported vocabulary comprehension and production, communication and language use.

Target vocabulary use and communication use

Language transcripts were created using the Systematic Analysis of Language Transcripts (SALT; Miller and Chapman, 1985) software program to measure the children's target vocabulary use during the intervention at sessions 18 and 24 and communication at baseline and sessions 18 and 24. Only parent–child interactions were used for transcript creation. Transcribers were masked to the specific AC intervention but it was not possible to mask the videos from the SC group since it was clearly visible that they did not have an SDG. For a more detailed description of how the transcripts were created, see Romski et al. (2010). Reliably trained transcribers coded videos of sessions 18 and 24 for spontaneous target augmented vocabulary use, defined in the original studies as a physical indication of target vocabulary symbol use on the SGD, and target spoken vocabulary word use, defined in the original studies as a combination of sounds that were consistently and meaningfully identified by the transcriber as a target word. When describing the number and proportion of target vocabulary used, only unprompted and non-imitative spoken and augmented words were counted. Functional vocabulary is the combined total of different spoken and augmented words used. The proportion of target vocabulary was calculated by dividing the number of target vocabulary used divided by the total vocabulary available for the child's use.

Once the transcripts were created, the SALT program provided six measures of child communication: (1) mean length of utterance in morphemes (MLU_m = [total number of morphemes] / [total number of utterances]), (2) mean length of turn ([total number of utterances] / [total number of turns]), (3) type/token ratio (TT ratio = [number of different words] / [total number of words used]), (4) utterance intelligibility (SALT defined utterance intelligibility as [number of intelligible words] / [number of total words]), (5) total turns (SALT defined total turns as one or more consecutive utterances), and (6) the

TABLE 3 Sequence of intervention sessions and parent role.

Week/ sessions	SLP role	Parent role in session		
		Play	Books	Snack
1/1,2	LAB – Parent observed the interventionist communicating with the child during the three activities as the SLP described the intervention to the parent and answered parent questions	O	O	O
2/3,4	LAB – Parent and SLP observed session run by interventionist	O	O	O
3/5,6	LAB – Parent and SLP observed session run by interventionist	O	O	O
4/7,8	LAB – Parent and SLP observed session run by interventionist	O	O	O
5/9,10	LAB – Parent and SLP observed session run by interventionist and parent joined the session for snack	O	O	X
6/11,12	LAB – Parent and SLP observed the session run by the interventionist and the parent joined the session for book + snack	O	X	X
7/13,14	LAB – Parent joined the entire session with the interventionist	X	X	X
8/15,16	LAB – Parent led the entire session; intervention supported parent as needed	X	X	X
9/17,18	LAB – Parent led the entire session; intervention supported parent as needed	X	X	X
10/19,20	HOME – Parent led the entire session; intervention supported parent as needed	X	X	X
11/21,22	HOME – Parent led the entire session; intervention supported parent as needed	X	X	X
12/23,24	HOME – Parent led the entire session; intervention supported parent as needed	X	X	X

O, observed; X, in session.

total number of spoken and/or augmented words used as coded on the transcripts.

Results

Table 4 reports means, standard deviations, and Mann–Whitney U results for the number and proportion of target vocabulary used in the form of spoken words, augmented symbols, and functional vocabulary. A non-parametric Mann–Whitney U test was run to determine if there were differences in children's target vocabulary between the AC and SC intervention groups during sessions 18 and 24. Significant differences were found between children in the AC and SC groups for the number of functional vocabulary targets used, the proportion of functional vocabulary used, and the total vocabulary targets provided during intervention sessions 18 and 24. At session 18, the number of functional vocabulary targets used by children in the AC group was significantly higher (mean rank = 18.00) than the number of functional vocabulary used by children in the SC group (mean rank = 3.50), $U = 138$, $z = 3.73$, $p = 0.000$. The proportion of functional vocabulary used for children in the AC group was significantly higher (mean rank = 18.00) than the proportion of functional vocabulary used by children in the SC group (mean rank = 3.50), $U = 138$, $z = 3.72$, $p = 0.000$. The total vocabulary targets provided was significantly higher for children in the AC group (mean rank = 17.15) than children in the SC group (mean rank = 6.75), $U = 118.50$, $z = 2.7$, $p = 0.005$. At session 24, the number of functional vocabulary items used by children in the AC group continued to be significantly higher (mean rank = 18.00) than the number of functional vocabulary used by children in the SC group (mean rank = 3.50), $U = 138$, $z = 3.72$, $p = 0.000$. The proportion of functional vocabulary used for children in the AC group was significantly higher (mean rank = 18.00) than the proportion of functional vocabulary used by children in the SC group (mean rank = 3.50), $U = 138$, $z = 3.72$, $p = 0.000$. The total number of vocabulary targets available for use was significantly higher for children in the AC group (mean rank = 17.22) than children in the SC group (mean rank = 6.50), $U = 120.00$, $z = 2.77$, $p = 0.004$.

Table 5 reports means, standard deviations and Mann–Whitney U results for parent and child communication measures of Mean Length of Utterance in morphemes (MLUm), Mean length of turn (ML turns), total turns, type-token ratio (TTR), and child communication intelligibility. A Mann–Whitney U test was run to determine differences in child and parent communication measures in the AC SC intervention groups during baseline, session 18, and session 24. There were no significant differences between the children in the AC and SC groups at baseline for any of the measures. At session 18, children in the AC group had a significantly higher TTR, % of intelligible communication utterances, and total number of turns. Parents of children in the AC group exhibited a larger MLUm and took significantly more turns than parents of children in the SC group. By session 24, children in the AC group had a significantly longer mean length of turns than children in the SC group. Children in the AC group continued to demonstrate a significantly higher percentage of intelligible communicative utterances. Parents of children in the AC group continued to use a significantly higher MLUm to communicate with their children than parents in the SC group.

Discussion

The children with DS who received AC intervention had stronger communication skills than the children who received the SC intervention as evidenced by significantly greater gains in functional vocabulary, and intelligible communication use. The AC interventions provided the children with a way to communicate via an SGD with visual-graphic symbols and speech output, while the children in the SC intervention were focused on producing intelligible spoken

TABLE 4 Comparing target vocabulary use between children with Down syndrome in augmented communication intervention and spoken communication intervention.

	Session 18					Session 24				
Variable	AC *n* =23	SC *n* =6	U	z	*p*	AC *n* =23	SC *n* =6	U	z	*p*
No. of augmented words used	11.74 (5.43)	–				12.3 (5.47)	–			
% of target augmented words used	0.60 (0.24)	–				0.63 (0.23)	–			
% of children using spoken words	0.52	0.33	82	0.81	0.51	0.39	0.33	73	0.26	0.85
No. of different spoken words	1.70 (3.88)	0.33 (0.52)	86	1.01	0.38	1.87 (4.77)	0.67(1.21)	74	0.31	0.81
% target spoken words	0.11 (0.10)	0.06 (0.00)	13	0.18	1.00	0.15 (0.14)	0.14(0.09)	9	0.00	1.00
functional vocabulary used	12.57 (6.31)	0.33 (0.52)	138	3.75	0.00**	12.82(6.51)	0.67(1.21)	138	3.73	0.00**
% functional vocabulary used	0.63 (0.22)	0.02 (0.03)	138	3.72	0.00**	0.64 (0.23)	0.05(0.08)	138	3.72	0.00**
Total target vocabulary available	19.83 (7.51)	15.17 (0.98)	118	2.69	0.01**	19.83(7.45)	15.17(0.98)	120	2.77	0.00**

Number of augmented and spoken words and percentages are means (with standard deviations in parentheses). *$p < 0.05$, two-tailed; **$p < 0.01$, two tailed.

vocabulary during the activities. The AC interventions did not hinder the children's spoken vocabulary development. Children in the AC intervention groups also maintained their gains in communication skills when the intervention transitioned from the lab environment to the home environment. Importantly, the findings from the current study suggested that the AC intervention did not hinder the spoken vocabulary development of the children with DS. Approximately half of the children with DS in the AC interventions produced spoken words by the end of intervention. In terms of utterance intelligibility, it is possible that children in the AC group may have a general advantage because words spoken with an SGD would be more intelligible than young children with only spoken word approximations.

These results are consistent with the broader findings from the two original studies that examined the parent-coached AAC interventions in a broader sample of children with developmental delays (Romski et al., 2010, 2023). These findings also support and enhance earlier case studies (e.g., Iacono and Duncum, 1995) that suggested SGDs may provide a viable communication intervention approach. It is also important to note that both the parents and children took more turns over time, which reflected a more balanced communicative exchange. It is important to note that the intervention is a combination of the use of an SGD with naturalistic communication strategies during established familiar routines. Overall, these findings suggest that SGDs are a viable intervention approach for young children with DS in the early stages of communication development. Furthermore, this study provides evidence that parents can learn to use SGDs with their children as young as 2 years of age when provided systematic coaching within naturalistic communicative exchanges.

There were some limitations to this study. First, although this study used data from two larger studies, the overall sample size of the children with DS who participated in the AC interventions was relatively small. The number of children who participated in the SC intervention was even smaller. Second, there was no comparison group of children with DS who received an AC intervention using an unaided form of AAC such as manual signs. It is not known how manual signs or PECS would have fared when compared to the AC interventions using SGDs. Additional studies are needed to carefully unpack the factors that compare the use of speech and both unaided (e.g., manual signs) and aided (e.g., SGD, PECS) forms of AAC. Third, the coding of the videos could not mask the SC vs. AC groups due to the inclusion of the SGD in the AC group interactions. It is important to note, however, that the coders were masked to the research questions and hypotheses of the studies. Fourth, parents were also taught to deliver naturalistic communication strategies as part of all the interventions. It is possible that the use of these strategies contributed to the children's increased vocabulary and the role the inclusion of these strategies created more opportunities for the children to produce vocabulary. This can not be ruled out but all interventions included the use of naturalistic communication strategies so they could account for differences between the AC and SC groups. Finally, at baseline, standard scores on the MSEL visual reception and receptive language domains were significantly higher among children who were randomly assigned to the augmented communication condition. This may suggest more developmental delays among children who were randomly assigned to the spoken communication condition, which may also have influenced study outcomes.

In conclusion, young children with DS can benefit from a parent-implemented augmented communication intervention that incorporates technology in the form of an SGD within naturalistic communicative routines. The children with DS who received the AC interventions had stronger communication skills at the end of the

TABLE 5 Comparing communication measures between children with Down syndrome in augmented and spoken communication intervention and their parents.

	Baseline					Session 18					Session 24				
Variable	AC *n* =23	SC *n* =6	U	Z	*p*	AC *n* =23	SC *n* =6	U	z	*p*	AC *n* =23	SC *n* =6	U	z	*p*
Child															
MLUm	0.97 (0.05)	0.99 (0.01)	57.5	−0.63	0.55	0.99 (0.06)	0.98 (0.03)	88	1.04	0.33	1.04 (0.13)	0.92 (0.12)	100	1.68	0.10
ML turns	1.11 (0.07)	1.19 (0.16)	54.50	−0.78	0.45	1.21 (0.14)	1.13 (0.14)	101	1.73	0.09	1.20 (0.14)	1.06 (0.04)	124	2.99	0.00**
TT ratio	0.05 (0.01)	0.04 (0.03)	90	1.14	0.28	0.12 (0.06)	0.05 (0.03)	121	2.81	0.00**	0.13 (0.07)	0.11 (0.08)	82.5	0.73	0.48
intelligibility	0.07 (0.07)	0.07 (0.10)	80	0.60	0.58	0.34 (0.18)	0.06 (0.07)	134	3.51	0.00**	0.42 (0.22)	0.12 (0.10)	124.5	2.99	0.00**
total turns	124.78 (72.33)	110.17 (42.22)	71	0.11	0.94	187.17 (74.95)	101.17 (37.76)	117	2.59	0.01*	162.87 (59.32)	102.50 (73.16)	101	1.72	0.09
Parent															
MLUm	3.31 (0.41)	3.09 (0.56)	75.5	0.35	0.73	3.41 (0.33)	3.03 (0.37)	108	2.10	0.04*	3.47 (0.39)	3.18 (0.26)	108	2.10	0.04*
ML turns	17.59 (23.06)	6.19 (3.32)	106	1.99	0.05	9.12 (6.98)	6.24 (2.09)	82	0.70	0.51	9.41 (6.85)	10.84 (10.06)	74.5	0.30	0.77
total turns	118.22 (65.99)	105.50 (41.05)	68.5	−0.03	0.98	168.48 (61.30)	97.83 (32.99)	119.5	2.72	0.00**	156.00 (54.23)	105.17 (70.86)	94.00	1.35	0.19

AC, augmented communication intervention; SC, spoken communication intervention; MLUm, mean length of utterance in morphemes; TT ratio, type token ratio; Intelligibility, % of intelligible utterances; ML turns, mean length of turn in utterances; Variable means are reported (with standard deviations in parentheses). $^{*}p < 0.05$, two-tailed; $^{**}p < 0.01$, two tailed.

24-session intervention than the children who received the SC intervention. The AC interventions provided the children with a means to communicate via an SGD. In contrast, the children in the SC intervention were still developing their use of spoken words. At the end of the intervention, children in the AC group had a larger functional vocabulary with which to communicate and were more intelligible than the children who received the SC intervention. The AC intervention did not hinder the children's spoken vocabulary development and in fact, was comparable, if not better than, the children's speech development in the SC intervention.

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.

Ethics statement

The studies involving human participants were reviewed and approved by Insitutional Review Board, Georgia State University, Atlanta, GA. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

MR, RS, and AB-H contributed to the conception and design of this study and wrote additional portions of the manuscript. AB-H, CW, EF, GK, and MK organized and compiled this database from the original studies. GK performed the statistical analyses. MR, AB-H, and PA wrote the first draft of the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Morphosyntactic development in German-speaking individuals with Down syndrome—longitudinal data

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Introduction: The present study provides longitudinal data on the development of receptive and expressive grammar in children and adolescents with Down syndrome and addresses the role of nonverbal cognitive abilities and verbal short-term memory for morphosyntactic development.

Method: Seventeen German-speaking individuals with Down syndrome (aged 4;6–17;1 years at first testing (T1)) were assessed twice, 4;4–6;6 years apart. For a subset of five participants, there was also a third assessment 2 years after the second. Receptive grammar, nonverbal cognition, and verbal short-term memory were tested using standardized measures. For expressive grammar, elicitation tasks were used to assess the production of subject-verb agreement and of *wh*-questions.

Results: At group level, the participants showed a significant increase in grammar comprehension from T1 to T2. However, progress diminished with increasing chronological age. Notable growth could not be observed beyond the age of 10 years.

With respect to expressive grammatical abilities, progress was limited to those participants who had mastered verbal agreement inflection around age 10 years. Individuals who did not master verbal agreement by late childhood achieved no progress in producing *wh*-questions, either.

There was an increase in nonverbal cognitive abilities in the majority of participants. Results for verbal short-term memory followed a similar pattern as those for grammar comprehension. Finally, neither nonverbal cognition nor verbal short-term memory were related to changes in receptive or expressive grammar.

Discussion: The results point to a slowdown in the acquisition of receptive grammar which starts before the teenage years. For expressive grammar, improvement in *wh*-question production only occurred in individuals with good performance in subject-verb agreement marking, which suggests that the latter might have a trigger function for further grammatical development in German-speaking individuals with Down syndrome. The study provides no indication that nonverbal cognitive abilities or verbal short-term memory performance determined the receptive or expressive development. The results lead to clinical implications for language therapy.

KEYWORDS

Down syndrome, language development, expressive grammar, receptive grammar, subject-verb agreement, *wh*-questions, verbal short-term memory, longitudinal study

1. Introduction

Down syndrome is one of the most common genetic disorders, caused by a third copy of chromosome 21 or part thereof (Martin et al., 2009; Loane et al., 2013). It is associated with both intellectual disability and language deficits. Language acquisition is overall delayed, but not all language domains are affected to the same extent. Individuals with Down syndrome often display severe impairments in the area of morphosyntax (for overview see Abbeduto et al., 2007; Roberts et al., 2008) as opposed to vocabulary skills or communicative and pragmatic competencies (Vicari et al., 2000; Grieco et al., 2015).

Receptive grammatical abilities are often considered to be less affected than expressive skills (Chapman et al., 1998), but many individuals with Down syndrome still exhibit difficulties in sentence comprehension (see review by Andreou and Chartomatsidou, 2020). Such difficulties often limit the comprehension of so-called non-canonical sentences in which the word order does not correspond to the unmarked constituent structure in a given language (e.g., passives or object-initial questions; Wimmer and Penke, 2020). They can, however, also affect the comprehension of syntactically simple sentences, such as simple active sentences (Witecy and Penke, 2017).

Sentence production can be limited to short utterances (Fowler et al., 1994) and is often characterized by frequent omissions of free and bound grammatical morphemes (Chapman et al., 1998). If longer utterances are produced, they can be incoherent and fragmental (Neitzel and Penke, 2021). Studies that used elicitation techniques to assess specific sentence structures revealed considerable difficulties with the production of syntactically complex sentences, for example different kinds of *wh*-questions (Tsakiridou, 2006; Joffe and Varlokosta, 2007; Wimmer et al., 2020). Studies analyzing spontaneous speech or data from elicitation or sentence repetition tasks also observed a particular deficit with the production of inflectional morphology in individuals with Down syndrome (e.g., Eadie et al., 2002; Laws and Bishop, 2003; Caselli et al., 2008; Penke, 2018). Most prominent are difficulties with verbal agreement and/or tense inflection that were found for different languages, such as English (e.g., Eadie et al., 2002; Laws and Bishop, 2003), Dutch (Bol and Kuiken, 1990), or German (Penke, 2018). In language production such affixes are often omitted or substituted by markers expressing different grammatical information (for instance, nonfinite markers; Penke, 2018).

Despite the existence of overarching symptoms that characterize the language difficulties in Down syndrome, individuals with Down syndrome exhibit large individual variability with respect to their language abilities. In many cases grammatical comprehension and production are impaired as described above, and some individuals with Down syndrome continue to show difficulties even with basic morphosyntactic structures into adolescence or adulthood (Fowler et al., 1994; Rondal and Comblain, 1996). Others, however, are able to comprehend or produce complex sentences (e.g., Thordardottir et al., 2002), and some individuals even appear to have nearly unimpaired language abilities (e.g., Rondal, 1995).

Besides the individual variability in language achievements in individuals with Down syndrome, a common finding is that the morphosyntactic development of individuals with Down syndrome is protracted and lags considerably behind chronological and often also mental age (e.g., Chapman et al., 1998). This raises questions regarding the timeframe in which morphosyntactic abilities are likely to develop further. In a cross-sectional study with 58 participants with Down syndrome, aged 4;6–40;3 years; Witecy and Penke (2017) found a positive correlation of grammar comprehension abilities with chronological age in children and adolescents (up to the age of 20 years), but not in adults. Similar results were reported by Facon and Magis (2019) and Iacono et al. (2010). Correlational analyses in these studies revealed a positive relation between chronological age and language comprehension measured through standardized tests in a group of children and adolescents ($n = 62$, aged 7–22 years; Facon and Magis, 2019), but not in adults ($n = 55$, aged 19–58 years; Iacono et al., 2010). Taken together, these findings suggest an ongoing development of receptive grammar in adolescence and subsequently, the building of a plateau in adulthood.

Rondal and Comblain (1996) conclude from their own and other cross-sectional data that grammatical development already ends around the age of 12–14 years in individuals with Down syndrome, not only in the receptive but also in the expressive domain. In contrast, based on the finding that the mean length of utterance (MLU) in narrative discourse increased with chronological age in a cross-sectional sample of 24 participants with Down syndrome (12;5–20;4 years), Thordardottir et al. (2002) assert that expressive syntactic development proceeds into late adolescence. Iacono et al. (2010)—in contrast—found a negative correlation between age and utterance lengths in narratives in their sample of adult participants.

Although these cross-sectional studies give some indications regarding the development of grammar, longitudinal data are needed to draw reliable conclusions on developmental trajectories in individuals with Down syndrome. The number of studies that have examined the development of grammatical abilities in individuals with Down syndrome longitudinally is, however, scarce, especially for expressive grammar. With respect to grammar comprehension, Conners et al. (2018) did not find significant growth (receptive grammar measured by the *Test for Reception of Grammar*, 2nd edition (TROG-2); Bishop, 2003) in a sample of 42 individuals with Down syndrome (chronological age 10–21 years at Time 1) over a period of 2 years. It is possible though, that progress in individuals with Down syndrome is slow and could therefore not be detected due to the relatively short time span that elapsed between the first and the second measurement (2 years later) in this study. Indeed, in a study by Laws and Gunn (2004) with two assessments 5 years apart, increases in grammar comprehension (measured by TROG scores) could be observed in most of the 30 participants (chronological age at study start: 5–19 years). However, younger participants made more progress than older ones and some of the latter even showed declining scores. Chapman et al. (2002) estimated individual growth functions for syntax comprehension (measured by the *Test of Auditory Comprehension of Language–Revised* (TACL-R), Carrow-Woolfolk, 1985) using hierarchical linear modelling. They collected data of 31 participants with Down syndrome who were tested four times over a period of 6 years (chronological age at first assessment: 5–20 years). The model predicted that there is still some progress in comprehension abilities in individuals aged around 12 years, but that the receptive skills are likely to decline in individuals aged 17 years or older, that is, in late adolescence and early adulthood.

Regarding expressive grammar, existing longitudinal evidence is conflicting as to whether there is improvement with increasing chronological age in adolescence or not. Support for the former comes from the study by Chapman et al. (2002) who estimated individual growth functions not only for receptive but also for productive syntactic abilities. In the four assessments over the span of 6 years, they found ongoing development in language production as measured by MLU, obtained in narrative samples, for most of the participants. Growth in expressive syntax over 2 years was also reported by Martin et al. (2013). They assessed expressive syntactic abilities using a standardized measure (Syntax Construction subtest of *Comprehensive Assessment of Spoken Language, CASL*; Carrow-Woolfolk, 1999) in up to three waves, each 1 year apart, in a varying number of boys with Down syndrome (Time 1: $n = 32$, Time 2: $n = 21$, Time 3: $n = 16$). Conners et al. (2018), on the other hand, did not observe progress in grammar production, neither in a standardized measure of expressive morphology (the word structure subtest of the CELF-P2; Wiig et al., 2004) nor in MLU based on narrations elicited by wordless picture books, a finding which conforms to their observation regarding receptive grammatical abilities. Again, as already mentioned above and as the authors state themselves, growth might have been so modest that it did not become apparent in the time span of 2 years elapsing between first and second testing of the participants in this study.

In summary, previous investigations point to a slowdown of the development in receptive grammar in adolescence which is followed by a plateau or even a decline. It remains unclear, however, whether the same applies to productive grammatical abilities or whether there is ongoing growth in adolescence. Furthermore, the question arises, which factors play a role in determining the course of development.

One factor that has been discussed to be associated with the language development in individuals with Down syndrome are nonverbal cognitive abilities or rather the limitations in this respect. Nonverbal cognitive abilities include visual–spatial processing and inductive reasoning skills. They are usually measured using standardized tests or subtests thereof that require only minimal or no verbal instructions (e.g., Leiter-R (Roid and Miller, 1997), Stanford-Binet 4th edition (Thorndike et al., 1986), Raven's Colored Progressive Matrices (Raven et al., 1995), Snijders-Omen Nonverbal Intelligence Test (Tellegen et al., 2007)). A number of cross-sectional studies have revealed a positive relationship between receptive or expressive grammatical abilities and nonverbal cognition in individuals with Down syndrome (Chapman et al., 1991; Abbeduto et al., 2003; Aktaş, 2004; Price et al., 2007, 2008; Iacono et al., 2010; Estigarribia et al., 2012; Finestack et al., 2013). That is, higher nonverbal cognitive abilities, assessed using the aforementioned measures, were correlated with higher grammatical abilities. Due to the fact that only one point in time is measured in these investigations, it is, however, unclear whether the observed relation indeed reflects a developmental association. Regarding the development of nonverbal cognitive abilities themselves, previous research has reported slowed, but continuing growth into adulthood (Couzens et al., 2011; Channell et al., 2014; Grieco et al., 2015). This ongoing development contrasts with the just presented studies on the development of grammatical abilities that have observed a standstill in receptive and/or expressive grammar (Chapman et al., 2002; Laws and Gunn, 2004; Conners et al., 2018). The question is whether this speaks against nonverbal cognition as a determining factor for the development of grammar or whether variation in nonverbal cognitive abilities still might be a predictor. First longitudinal evidence for the former view is provided by Chapman et al. (2002). They did not find nonverbal cognition, as measured by the Pattern Analysis subtest from the Stanford-Binet 4th edition, which assesses visual–spatial processing (Thorndike et al., 1986), to predict individual growth in grammar comprehension or production. In Conners et al. (2018) and Martin et al. (2013) nonverbal cognitive ability was included as a covariate in data analysis, but its role as a predictor for grammar development was not explicitly analyzed. We are not aware of any other longitudinal studies that have examined the relation between nonverbal cognitive abilities and grammar development to this date. Thus, further research in this respect is needed.

Apart from nonverbal cognition, morphosyntactic development could be influenced by weak verbal short-term memory skills that constitute another characteristic symptom in individuals with Down syndrome (*cf.* meta-analyses by Næss et al., 2011; Godfrey and Lee, 2018). According to Baddeley's influential multicomponent model on working memory (e.g., Baddeley, 2003; Baddeley et al., 2021), verbal short-term memory represents the memory component most relevant for language. It comprises a passive capacity-restricted phonological store that maintains phonological information (e.g., words and sentences) for up to 2 s. Memory traces can be refreshed by a rehearsal process, a kind of inner speech. The crucial role of this phonological loop component consisting of storage and rehearsal is to enable the hearer to extract the relevant morphosyntactic information from the speech signal during processing, a prerequisite to language comprehension and grammar development. Verbal short-term memory skills have been shown to play an important role in typical and atypical acquisition of morphosyntax (*cf.* recent papers by Delage and Frauenfelder, 2020; Roesch and Chondrogianni, 2021).

In most studies on Down syndrome that relate morphosyntactic comprehension or production skills to the performance in verbal short-term memory tasks, significant relations between the two domains have been found (e.g., Laws and Bishop, 2003; Laws and Gunn, 2004; Miolo et al., 2005; Estigarribia et al., 2012; Frizelle et al., 2019b; Wimmer et al., 2020). However, the majority of these studies are cross-sectional and the observed relations might also be due to task demands. This might especially hold for grammar comprehension which is usually assessed using sentence picture-matching tasks that place high demands on verbal short-term memory (*cf.* Frizelle et al., 2019a; Penke and Wimmer, 2020 for discussion). Longitudinal studies are rare so far, but existing studies have found verbal short-term memory capacity—measured by nonword repetition—to be a predictor for progress in grammar comprehension (Chapman et al., 2002; Laws and Gunn, 2004). However, in the investigation by Laws and Gunn (2004), this only held for the younger participants that were aged below 10 years at initial assessment. This suggests that verbal short-term memory may play an important role for the acquisition of receptive grammar, especially in childhood and early adolescence, but that it might be less relevant for progress of language abilities in older individuals with Down syndrome.

Whether verbal short-term memory capacities are also associated with the development of expressive morphosyntactic abilities in individuals with Down syndrome, is unclear yet. In the study by Chapman et al. (2002), performance with respect to verbal short-term memory did not predict development in expressive grammar (as measured by MLU). However, growth in expressive grammar was predicted by abilities in grammar comprehension which in turn were related to short-term memory capacity. Hence, there might be an indirect relation between the latter and productive grammatical abilities mediated by comprehension abilities.

To summarize, the following open issues arise from the current state of research: Whereas existing studies reveal a clear tendency for the developmental course in grammar comprehension, namely, a levelling off of the development in adolescence, the picture is less clear for expressive morphosyntactic abilities. Furthermore, more longitudinal research is needed that targets the role of nonverbal cognition and verbal short-term memory as potential influencing factors for morphosyntactic development in production and comprehension. Against this background, the aim of our study is to investigate both grammar comprehension and production as well as the described potential predictor variables longitudinally in the same sample of individuals with Down syndrome.[1] In doing so, we will not only look at the performance of the group, but we will focus on individual development. Explicit investigations of individual differences have often been neglected in previous studies on Down syndrome, but seem important given the reports of large inter-subject variability in the literature (Conners et al., 2018).

Difficulties in grammar comprehension or production can negatively affect the communication and participation of individuals with Down syndrome, as they may be less able to follow conversations and prompts in their environment or to express their needs and thoughts. This is particularly true in educational or employment settings. In addition, impaired grammar comprehension can also significantly impede intervention in other language areas, such as vocabulary or expressive morphosyntax. Thus, understanding both the nature of the receptive and expressive grammatical difficulties and their developmental course is important for practitioners in therapeutic as well as educational and vocational contexts.

The research questions of the current investigation are: (RQ1) What is the course of development in (a) receptive and (b) expressive morphosyntactic abilities in individuals with Down syndrome? (RQ2) What is the role of nonverbal cognition and verbal short-term memory in determining the developmental progress in morphosyntax?

2. Materials and methods

2.1. Participants

Seventeen German-speaking individuals with Down syndrome (7 female, 10 male; chronological age at study start: 4;6–17;1 years) were assessed twice, 4;4–6;6 years apart.[2] Information on the chronological ages of the participants at the different points of assessment are presented in Table 1 (see Supplementary material for individual data). The nonverbal mental age of the participants ranged between 3;5 and 6;5 years at the initial assessments (T1) (*M* = 4;8, *SD* = 1;0). Participants were included in the study if they were monolingual German speakers, used oral language as their primary means of communication, and produced at least two-word-utterances. This was confirmed by the parents, and the latter two aspects were additionally checked during the first assessment session. At T1 participants' parents were asked to fill out a questionnaire to provide information on biographical and medical background, including questions on hearing and ear infections, kindergarten/school attendance, speech and language therapy, and their own level of education. At the second testing (T2), a further questionnaire was given to follow up on part of these aspects. All children, adolescents, and young adults in the present study had normal or corrected vision as well as normal hearing, with the exception of one participant who was reported to have a mild conductive hearing loss of 35 dB in one ear. The participants all attended inclusive kindergartens or inclusive or specials needs schools at T1 and were still in school at T2, apart from one young adult who was working in a sheltered employment facility at the time of the second assessment. To gain a more comprehensive picture of the development of expressive grammatical abilities, we tested those five individuals of the initial cohort of 17 participants a third time (T3) who had not mastered the grammatical structures under study at T2. This third testing took place 2 years after the second assessment and 8 years after the first (see Table 1).

2.2. Measures

2.2.1. Language measures (RQ1)

2.2.1.1. Receptive grammar: TROG-D

The standardized measure TROG-D (Fox, 2011), a German adaption of the Test for Reception of Grammar (Bishop, 1983, 2003), was applied to assess grammar comprehension. The TROG is widely used in research of language development in different populations and languages as well as in clinical practice. It has also been employed in the longitudinal studies by Laws and Gunn (2004) and Conners et al. (2018) to assess receptive grammatical abilities in individuals with Down syndrome. The internal consistency reliability (Cronbach's α) of the TROG-D is 0.90. The TROG-D is correlated with the sentence comprehension subtest from the SETK 3-5 (Sprachentwicklungstest für drei- bis fünfjährige Kinder "Test of language development for three to five year old children"; Grimm, 2001) at $r = .72$ (Fox, 2011).

Participants were verbally presented with a word or a sentence and had to choose the corresponding picture out of a choice of four. The test includes 21 blocks of four items each. Each block tests a different grammatical structure which increases in grammatical complexity. In accordance with the manual, testing was discontinued when the participant gave at least one incorrect answer in five consecutive blocks. Raw scores were used for the analyses. They result from the number of blocks, in which all items have been answered correctly, and therefore might range between 0 and 21 points.

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[1] Note that data on grammar comprehension as well as predictor variables has been previously published in a German paper for a German-speaking readership of speech and language therapists (Witecy et al., 2021).

[2] The participants were part of a larger sample that was first examined in a cross-sectional study on the grammatical abilities of children and adolescents with Down syndrome. The initial cohort included 31 children and adolescents. While the initial assessment of this cohort was funded by the German Science Foundation, the follow-up assessments reported in this paper could only take place as personnel, financial and time resources permitted. This accounts for the protracted assessment period at T2. Furthermore, data collection was delayed because of COVID-19 related regulations. Participants of the initial cohort, respectively their parents, were contacted for assessment at T2. However, only 20 families agreed to further participation of which one family had moved too far away and two participants could not be assessed due to increased COVID-19 related restrictions. This resulted in a sample of 17 participants for the follow-up.

TABLE 1 Overview of participants (ages in years; months).

N	Sex	Chronological age at T1	Chronological age at T2	Time span from T1 to T2
17	7 female 10 male	Range: 4;6–17;1 *M* = 9;10 (*SD* = 3;3) *Mdn* = 9;6	Range: 11;0–23;2 *M* = 15;7 (SD = 3;3) *Mdn* = 15;5	Range: 4;4–6;6 years *M* = 5;9 (*SD* = 0;8) *Mdn* = 6;0
Retested at T3				Chronological age at T3
5	2 female 3 male	Range 4;6–12;0 *M* = 8;8 *Mdn* = 8	Range: 11;0–18;1 *M* = 14;9 *Mdn* = 13;8	Range: 13;0–20;0 *M* = 16;8 *Mdn* = 15;7

2.2.1.2. Expressive grammar: elicitation tasks on subject-verb agreement and *wh*-question production

Previous studies on productive grammatical abilities have employed MLU as a measure indicating grammatical complexity. MLU provides an indirect measure of morphosyntactic development, the assumption being that the longer the utterance the more complex the structure. For individuals with Down syndrome, however, this assumption does not seem to hold. In an investigation of narrations produced by German individuals with Down syndrome, we found high MLU values to often come about by ungrammatical concatenations of sentence fragments within one utterance (Neitzel and Penke, 2021). Here, we therefore adopted two elicitation tasks to assess expressive grammatical abilities more directly: one focusing on verbal agreement inflection and the other on *wh*-question production. Both phenomena have been found to often be affected in individuals with Down syndrome (e.g. Penke, 2018; Wimmer et al., 2020) and the two tasks have been applied successfully to assess these phenomena in individuals with language impairments in the past.

A video description task was performed to elicit verb forms marked for subject-verb agreement. Participants had to describe the action depicted in 30 short, silent video scenes presented on a laptop computer. They were prompted by the question *Was passiert da?* "What is happening here?". In the videos the participants could either see the experimenter herself, a single child, or two children performing an action and were therefore expected to produce verbs inflected for 2nd person singular (e.g., *du schreib-st* "you are writing" for videos showing the experimenter), 3rd person singular (e.g., *er koch-t* "he is cooking" for videos showing a single child), or 3rd person plural (e.g., *sie lauf-en* "they are running" showing two children). First, participants were familiarized with the task by three practice items in which the acting characters were introduced. Subsequently, there were 10 target videos for each grammatical context (2nd person singular, 3rd person singular, 3rd person plural). All 30 target videos were presented in a previously fixed randomized order. Accuracy scores for correct agreement inflection were determined for all utterances that consisted of both an overt subject and a main verb. An utterance was scored as correct if the suffix on the verb agreed with the subject. Both unmarked verbs and substitutions of the ending were considered incorrect.[3]

In addition, we assessed the production of complex syntactic structures by eliciting *wh*-questions. We collapsed data that came from two methodically comparable tasks eliciting *wh*-questions (see Table 2). In both tasks, participants were instructed to pose different *wh*-questions to either a toy figurine or to toy animals (e.g., "Ask the snail what it is doing."; see Table 2 for more details on the item material). At T1 only Task 1 was used.[4] At T2 six participants were administered Task 1 and seven were assessed using Task 2. As the structure and content of the questions as well as the method of elicitation in a playful setting and the number of questions are comparable in the two tasks, we combined the data at T2. Similar question elicitation tasks with puppet scenarios or pictures are common and adequate tools to evaluate expressive grammatical abilities in children (*cf.* Thornton, 1996). They have been used successfully in the past in children with developmental language disorders of diverse etiology (for Down syndrome and Williams syndrome, e.g., Joffe and Varlokosta, 2007, for children with Autism Spectrum Disorder and children with Developmental Language Disorder *cf.* Sukenik et al., 2021).

Accuracy scores for *wh*-question production were determined for all utterances that contained an overt *wh*-element and/or displayed a clear raising question intonation. Questions were judged as syntactically correct when there was a fronted *wh*-word, a finite verb in second position, and a subject (which could also be the *wh*-word).

2.2.2. Cognitive measures as potential predictors for grammatical development (RQ 2)

As we were not only interested in the development of the receptive and expressive grammatical abilities but also wanted to evaluate whether nonverbal cognition and verbal short-term memory play a role in determining the development in these areas, the following measures were included to assess these variables.

2.2.2.1. Nonverbal cognition: reasoning scale of the SON-R 2.5–7

The Reasoning Scale of the Snijders-Omen Nonverbal Intelligence Test (SON-R 2.5–7; Tellegen et al., 2007) was used to

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[3] For a more detailed description of the task and cross-sectional results for a larger sample, which includes the participants in the current study, see Penke (2018).

[4] Further information on the task and detailed analyses of *wh*-question production in individuals with Down syndrome can be found in Wimmer et al. (2020).

TABLE 2 Overview of task and item material for *wh*-question production tasks 1 and 2.

	Wh-questions task 1	*Wh*-questions task 2
n items overall	*n* = 14	*n* = 12
instrument	'Ask the snail' game (*cf.* Wimmer et al., 2020)	Subtest 1 of ESGRAF 4–8, Item 1–12 (Motsch and Rietz, 2016)
task	Pose questions to a figurine (snail/robot) (structured dialogue)	Pose questions to identify three toy animals (monkey, pig, goose) hidden in a box
instruction	Frag die Schnecke, was sie hier macht. (“Ask the snail what it is doing.”)	Frag das Tier, was es fressen mag. (“Ask the animal what it would like to eat.”)
wh-argument questions	*n* = 8 *wh*-subject and-object questions (*who/what* questions)	*n* = 6 *wh*-object questions (*what* questions)
example	target: Was machst du hier? (“What are you doing here?”)	target: Was magst du fressen? (“What do you like to eat?“)
wh-adjunct questions	*n* = 6 (*where/when/how* questions)	*n* = 6 (*where/how* questions)
example	Wo wohnt die Oma? (“Where is the grandma living?“)	Wo wohnst du? (“Where are you living?“)
Example of dialogue / instruction (in italics)	Examiner: Die Schnecke spricht leider nicht mit Erwachsenen, nur mit Kindern. *Frag die Schnecke, was sie hier macht.* (“Unfortunately, the snail does not speak to adults. *Ask the snail what it is doing.*”) Child: Was machst du hier? (“What are you doing here?”) Examiner (takes the role of the snail): Ich besuche jemanden. (“I am visiting someone.”)	Examiner: Das Tier ist in der Box versteckt. *Frag das Tier, was es fressen mag.* („The animal is hidden in the box. *Ask the animal what it would like to eat.*”) Child: Was magst du fressen? (“What do you like to eat?“) Examiner (takes the role of the hidden animal): Ich fresse gern Bananen. (“I like to eat bananas.”)

assess nonverbal cognitive abilities. It consists of three subtests (Categories, Analogies, Situations) that test concrete and abstract reasoning skills. It is normed for the ages of 2;6 to 7;11 years. Reported internal consistency of the Reasoning Scale is $r = 0.83$. Validity is confirmed by a high correlation ($r = 0.74$) with the nonverbal scale of the K-ABC (Melchers and Preuß, 2001; Tellegen et al., 2007). Total raw scores were used in the analyses.[5] In addition, nonverbal mental age equivalents were computed to describe the sample.

2.2.2.2. Verbal short-term memory: nonword repetition subtest of the SETK 3–5

A common task to assess verbal short-term memory is the repetition of nonwords. It is also well-suited for individuals with Down syndrome and has been used frequently in previous investigations (e.g., Laws and Gunn, 2004; Conners et al., 2018). In the current study, the nonword repetition subtest of the SETK 3–5 (Grimm, 2001) was employed. It comprises 18 nonwords with a length of two to five syllables. The nonwords were read to the participants, who had to repeat them accurately immediately after presentation. Raw scores, i.e., the number of correctly repeated nonwords, were used in the analyses (max. 18 points). Internal consistency reliability ranges between 0.73 and 0.81, depending on the age band (Grimm, 2001). Correlations with other measures of verbal short-term memory to provide information about validity are not reported.

2.3. Procedure

Data collection at T1 took place between 2013 and 2015 either in a quiet room at the university or at participants' homes. A broad range of language and cognitive measures, both experimental and standardized, was administered in four sessions (40–60 min). Here, we report only those measures which were repeated at second testing (T2). Testing at T2 was carried out between 2018 and 2020 and took place at participants' homes or in institutions for language therapy. A subgroup of five individuals was tested again at T3. Testing at T3 took place in 2022 and only included the measures of expressive syntax. Table 3 presents an overview on which tests were conducted at T1, T2, and T3.

In all testing sessions sufficient time for pause was given. The order of presentation of the different measures was usually the same, with an alternation of receptive and expressive tasks where possible. Standardized measures were applied according to the procedure described in the manuals. Testing took place after a time of familiarization of the participant with the examiner and the situation. Where possible, parents were not present during the testing sessions to avoid distraction.

Each session was audio- and videorecorded. Recordings were used for the transcription of participants' verbal responses or to check the scoring of participants' nonverbal responses in the tests on receptive grammar and nonverbal cognition. Responses in the tasks on subject-verb agreement and *wh*-question production were transcribed by a primary transcriber and transcripts were checked by a secondary transcriber. If necessary, disagreement was resolved with the assistance of a third qualified person. Utterances for which no interrater agreement could be achieved were not included in the analyses.

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[5] Performing the analyses with nonverbal mental age does not change the results.

Approval for data collection was obtained by the Ethics Committee of the Medical Department of the University of Cologne (numbers of approvals: 12-033, 18-121). Informed written consent was given by the parents or legal guardians of all participants and verbal consent from the participants was obtained on the test date.

2.4. Analyses

As raw scores are measured on an ordinal scale, non-parametric procedures were chosen.

2.4.1. Analyses addressing RQ1a,b

To find out if there was a significant group change between T1 and T2 in the different measures, Wilcoxon signed rank tests were computed. For the examination of individual change, difference scores for each participant and variable were determined by subtracting the result achieved at T1 from the result achieved at T2. Positive scores show progress, whereas negative scores indicate a decline in performance (see Table 4 for means, standard deviations, and range). To gain more insight into the time course of the development, we determined whether change in language measures was related to the chronological age of the participants. Therefore, we calculated Spearman's correlations between individual difference scores and chronological age at T1. To gain a more comprehensive picture of the development of expressive grammatical abilities, we performed a post-hoc exploratory analysis of the data for the expressive tasks where we looked for implicational relationships between the tested phenomena.

2.4.2. Analyses addressing RQ2

Analogous to the procedure for RQ1, we first examined group changes in the measures of nonverbal cognition and verbal short-term memory themselves using Wilcoxon signed rank tests. Individual changes were further explored descriptively. To investigate the relation between changes in receptive or expressive morphosyntactic abilities (difference scores) and nonverbal cognition (reasoning scale raw scores) or verbal short-term memory (nonword repetition scores) as possible influencing factors correlational analyses were performed for these measures.

3. Results

An overview of the results for T1 and T2 and the difference scores can be found in Table 4. Individual test results achieved at T1, T2, and T3 can be found in the Supplementary material to this text. No overall floor or ceiling effects could be observed in the measures used in this study.

3.1. Receptive grammar (RQ1a)

There was a significant increase in overall TROG-D raw scores between T1 and T2 ($z = 3.195$, $p = 0.001$, $r = 0.775$; *Mdn* T1 = 7, *Mdn* T2 = 9). However, difference scores were negatively correlated with chronological age at T1 [$r_s(15) = -0.717$, $p = 0.001$], indicating that older participants exhibited less growth in TROG-D scores than younger participants. The individual data obtained at T1 and T2 are presented in Figure 1. To obtain more information on the time course

TABLE 3 Number of participants that were tested at T1, T2, and T3.

	Nonverbal cognition	Verbal short-term memory	Receptive grammar	Expressive grammar		
Testing	Reasoning scale of the SON-R	Nonword repetition	TROG-D	Subject-verb agreement	*wh*-questions task 1	*wh*-questions task 2
T1	17	17	17	17	14	–
T2	17	16	17	5	6	7
T3				5	5	–

TABLE 4 Means (standard deviations) and ranges for first (T1) and second testing (T2) as well as difference scores (T2 minus T1).

		Time 1			Time 2		Difference	
	N T1	Mean (*SD*)	Range	*N* T2	Mean (*SD*)	Range	Mean (*SD*)	Range
Nonverbal mental age in months	17	56.2 (12.0)	41–77	17 (15)[a]	68.3[a] (26.1)	44–>96[a]	14.3[a] (11.4)	(−7)–29[a]
Reasoning scale raw scores	17	24.8 (5.7)	17–33	17	31.0 (6.4)	19–42	6.2 (4.3)	(−2)–12
Nonword repetition raw scores	17	6.1 (3.7)	0–13	16	7 (3.1)	1–11	1.3 (2.6)	(−3)–8
TROG-D raw scores	17	6.1 (2.7)	3–11	17	8.9 (2.8)	4–16	2.8 (2.5)	0–7
Subject-verb agreement accuracy scores	17	65.4% (28.4)	16.7–100%	5	85.3% (18.1)	44.4–100%	19.8 (19.4)	(−6.3)–54.9%
wh-production accuracy scores	14	58.61% (36.0)	0–100%	9	81.0% (30.8)	10–100%	25.6% (24.2)	0–80%

[a] The nonverbal mental age of two participants at T2 could not be determined exactly because their performance exceeded the norming sample of the SON-R 2.5–7. It can therefore only be estimated as at least 8;0 years (96 months). These participants were not included in the calculation of the mean mental age at T2 and the difference scores for mental age.

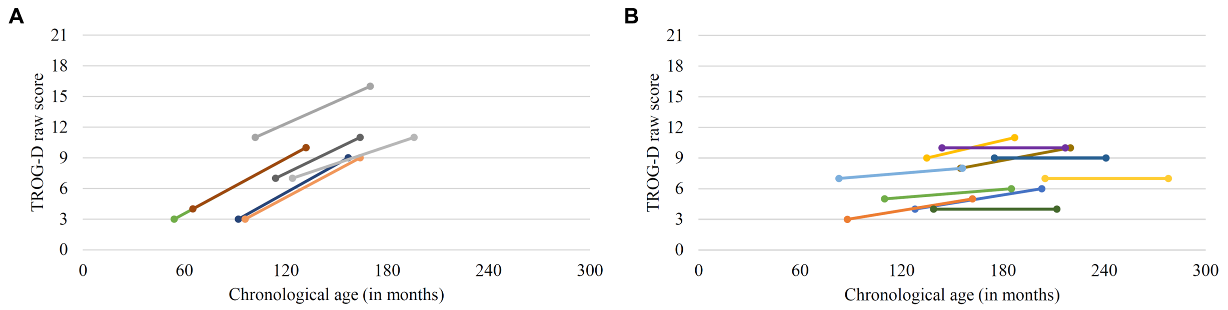

FIGURE 1
Individual change in Test for Reception of Grammar (TROG-D) raw scores. **(A)** Participants with notable change (difference scores ≥3) **(B)** Participants with little or no change.

of the development, individual change was further explored descriptively. The data show that a notable increase in raw points only occurred in individuals with a chronological age of 10 years or younger at T1. Since statistical measures to analyze individual change were not available for our data, an increase of three or more raw points was considered as notable. The oldest participant that achieved an improvement of three or more raw points in this test was aged 10;4 years at T1 (see Supplementary material). In contrast, for participants that were older than 10 years at T1, raw scores changed little or not at all (see Figure 1; Supplementary material).

3.2. Expressive grammar (RQ1b)

3.2.1. Subject-verb agreement

The majority of the sample (12 out of 17 individuals) already performed well at subject-verb agreement at T1 and achieved accuracy scores of over 85% (range 86.2–100%, mean 94.6%), leaving only five participants with the potential to improve notably by T2. The development of these five individuals was followed up on and is presented here and in Table 4. Also, due to the small number of data points at T2, we did not perform statistical tests. Of the five individuals that displayed problems with subject-verb agreement at T1 (range of accuracy scores 25–54.2%, mean 40.8%) three were younger and two older than 10 years (chronological age). All five individuals showed an improvement in accuracy scores at T2 (mean 70.5%, range 44.4–82.1%). To further investigate the progress of this group of individuals, the group was retested again at T3. However, further progress to accuracy scores of over 90% could only be determined for two of the five tested individuals, both younger than age 10 years at T1 (see data for P1 and P6 in the Supplementary material). For the other three individuals (P4, P13, and P14) no further progress occurred, instead accuracy scores declined from a mean score of 63% at T2 to a mean score of 51.6% at T3 (range of accuracy scores at T3 25–65.5%).

3.2.2. *Wh*-question production

The within-group comparison for *wh*-question production is based on the data of nine participants. Five participants achieved high accuracy scores (over 90%, range 90.9–100%) already at T1 and, therefore, displayed only a limited potential for improvement at T2. For three other individuals, it was not possible to perform the question elicitation task at T1 due to lack of cooperation, insufficient understanding of the task, and/or insufficient language skills. Thus, a difference in accuracy scores to T2 could not be determined. The comparison of T1 and T2 performance for the nine participants indicates significant growth between the two assessments in the group as a whole ($z = 2.666$, $p = 0.008$, $r = 0.889$; *Mdn* T1 = 45.45%, *Mdn* T2 = 85.71%). The mean accuracy score of 37.6% at T1 (range 0–71.4%) increased to 72.5% at T2 (range 10–100%). An inspection of the individual data reveals that seven of the nine individuals achieved a considerable improvement in accuracy scores for *wh*-question production between T1 and T2 (see Figure 2). For two individuals (P4 and P14), however, accuracy scores still were below 20% at T2, indicating a clear deficit in expressive grammatical abilities that persisted at T2. The correlation between *wh*-question production difference scores and chronological age at T1 did not yield a significant result [$r_s(7) = -0.317$, $p = 0.406$]. Thus, for the tested nine children and adolescents, the improvement in *wh*-question production seemed to be independent of their chronological age.

To determine the development in producing *wh*-questions in the three individuals who could not perform the task at T1 (P1, P6, and P13) and the two individuals who failed to produce *wh*-questions at T1 and only achieved minimal improvement at T2 (P4, P14), we retested *wh*-question production for these participants at T3. Two of the participants, who could not perform the task at T1 (P1 and P6), achieved accuracy scores of 38% and 58% at T3, indicative of an improvement in producing syntactically correct *wh*-questions. For the other three individuals (P4, P13, and P14), no substantial increase in accuracy scores could be observed at T3, and accuracy scores remained very low (range 0–18%).

3.2.3. Exploratory analysis for expressive grammar

With regard to their expressive morphosyntactic abilities the participants fall into three different groups. The first group, consisting of five individuals (P7, P8, P12, P15, and P16), achieved accuracy scores of over 80% in both expressive measures, indicating good performance with subject-verb agreement marking *and* the production of *wh*-questions already at T1.

The second group of seven individuals (P2, P3, P5, P9, P10, P11, and P17) had already obtained high accuracy scores of over 80% for subject-verb agreement at T1 while accuracy scores for *wh*-question production were lower at T1 and only reached 80% or more at T2, indicating that the development of *wh*-question production proceeded after subject-verb agreement marking had been mastered.

The third group of five individuals (P1, P4, P6, P13, and P14) obtained relatively low scores in both expressive grammatical measures at

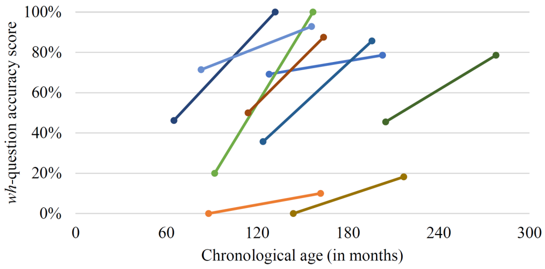

FIGURE 2
Individual change in *wh*-question production accuracy scores between T1 and T2.

T1 (range of accuracy scores for subject-verb agreement at T1 25–54.2%, accuracy scores in *wh*-question production at T1 0% if test could be performed). At T2 and T3, two of these individuals (P1 and P6) achieved accuracy scores of over 80% for subject-verb agreement while the accuracy scores for *wh*-question production did not reach this level at T3 (58.3 and 38.5%). For the other three individuals, accuracy scores for verbal agreement marking surpassed the accuracy scores for *wh*-question production at T2 and T3. However, none of these individuals achieved an accuracy score of 80% or above for either of the two expressive morphosyntactic measures at T3. For all three testing times, the data, thus, yield that progress in subject-verb agreement marking precedes progress in the production of *wh*-questions. The data do not contain a single case where an individual achieved good performance in the production of *wh*-questions but was impaired in the marking of subject-verb agreement.

This order of difficulty between performance in subject-verb agreement and production of *wh*-questions was confirmed by an implicational scaling analysis of the accuracy scores obtained by the participants for subject-verb agreement and for *wh*-question production at T1 (Guttman, 1944; Hatch and Lazaraton, 1991). The analysis was conducted to determine whether the acquisition of subject-verb agreement and the acquisition of *wh*-questions (both defined by an accuracy score of over 80%) display a scale, indicating that acquisition of the one phenomenon truly precedes acquisition of the other. As the data do not contain a single case where participants display better performance with respect to the production of *wh*-questions compared to the production of subject-verb agreement marking, the implicational analysis gave a perfect *Guttman coefficient of scalability* (=1). The coefficient allows to predict with 100% accuracy that an individual displaying good performance (>80% accuracy) with respect to the production of *wh*-questions will also achieve good performance with the marking of subject-verb agreement. The analysis, thus, implies a true developmental scale according to which acquisition of subject-verb agreement marking precedes the production of *wh*-questions.

3.3. Potential predictors for grammatical development (RQ2)

3.3.1. Nonverbal cognition

There was a significant increase in reasoning scale raw scores in the overall group ($z = 3.364$, $p < 0.001$, $r = 0.816$; *Mdn* T1 = 24, *Mdn* T2 = 31). On an individual level, raw scores increased for all participants except for two, who showed a decrease of 2 raw points or no change. Difference scores for the others ranged between 1 and 12. There was no significant correlation of reasoning scale difference scores with chronological age [$r_s(15) = -0.417$, $p = 0.096$], indicating that the improvement in nonverbal cognition was independent of participants' chronological age at T1.

3.3.2. Verbal short-term memory

There was no significant growth in nonword repetition scores in the group as a whole between T1 and T2 ($z = 1.870$, $p = 0.062$, $r = 0.468$; *Mdn* T1 = 7, *Mdn* T2 = 8). Note that one participant was missing a score at T2 as the test could not be performed due to lack of cooperation. Nonword repetition difference scores correlated negatively with chronological age at T1 [$r_s(14) = -0.566$, $p = 0.022$]. This indicates that older participants at T1 displayed less growth or even a decline in nonword repetition performance compared to younger participants. Figure 3A shows that participants with a notable change in nonword repetition scores (here defined as an increase in raw scores of three or more raw points) were younger than 10 years at T1 (chronological age). In contrast, participants that were older than 10 years at T1 displayed little or no change in raw scores for nonword repetition (Figure 3B). The oldest participant that achieved an improvement of three or more points in this task was 9;6 years at T1.

3.3.3. Relation of language change and nonverbal cognition or verbal short-term memory

The results of the correlational analyses that were performed to investigate the relation between language difference scores and performance on the reasoning scale of the SON-R or nonword repetition performance at T1 are displayed in Table 5. Chronological age was controlled in the correlations with reasoning scale raw scores as both were positively related [$r_s(15) = 0.644$, $p = 0.005$]. Analyses yielded that the difference scores obtained for receptive grammar (TROG-D difference scores) correlated neither with reasoning scale raw scores, measuring nonverbal cognition, nor with nonword repetition performance, our measure for verbal short-term memory capacities.

For expressive grammar, correlational analyses were only performed for the accuracy difference scores obtained for *wh*-question production from those nine individuals that were tested at T1 and T2. *Wh*-question production difference scores did not correlate with reasoning scale raw scores or nonword repetition scores at T1. For subject-verb agreement, no correlations between accuracy difference scores and SON-R reasoning scale or nonword repetition were computed, since the number of data points at T2 was too small (only five individuals). Note however, that the three participants who displayed no progress with respect to verbal agreement marking (P4, P13, and P14) nevertheless progressed with respect to their nonverbal cognitive abilities (increases in raw scores of 4, 8, and 9 points). This suggests that progress in nonverbal cognitive development is not linked to progress in the production of verbal agreement markings.

4. Discussion

4.1. Development of receptive and expressive morphosyntactic abilities (RQ1)

The main purpose of our study was to investigate the developmental course in receptive and expressive morphosyntactic abilities in individuals with Down syndrome. To this end, we analyzed data from 17 individuals

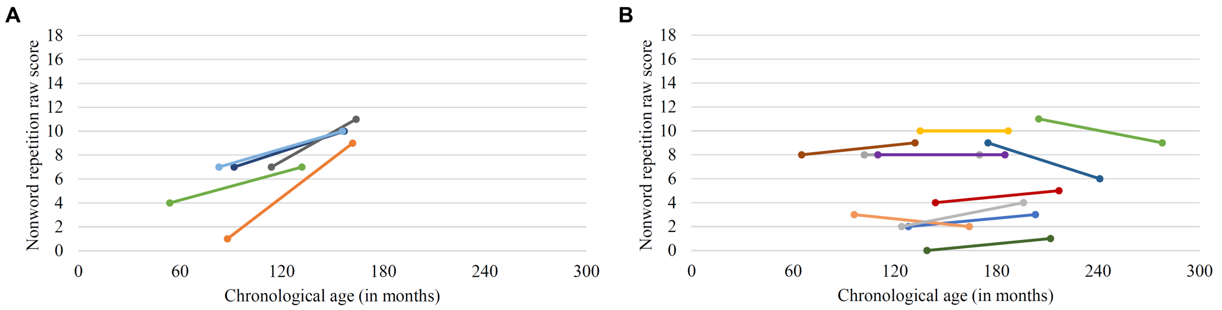

FIGURE 3
Individual change in nonword repetition scores. **(A)** Participants with notable change (difference scores ≥3) **(B)** Participants with little or no change or decline.

TABLE 5 Correlations between chronological age, reasoning scale raw scores, and nonword repetition scores at T1 and TROG-D and *wh*-question difference scores.

	Difference scores	
	TROG-D	*wh*-questions
Chronological age at T1	$r_s = -0.717$	$r_s = -0.317$
	$p = 0.001$	$p = 0.406$
	$n = 17$	$n = 9$
Reasoning scale raw scores at T1	$r_s = -0.146$	$r_s = 0.313$
	$p = 0.589$	$p = 0.451$
	$n = 17$	$n = 9$
Nonword repetition scores at T1	$r_s = -0.122$	$r_s = 0.545$
	$p = 0.640$	$p = 0.129$
	$n = 17$	$n = 9$

Correlations with reasoning scale raw scores were controlled for chronological age.

with Down syndrome that were collected at two time points about 4½–6½ years apart. For a subset of five participants, there was also an additional third assessment 2 years after the second.

Regarding grammar comprehension, we found a significant improvement between T1 and T2 in the group as a whole. However, this did not apply to all participants. A negative correlation of TROG-D difference scores with chronological age at T1 showed that there was less improvement in older individuals, suggesting a levelling off in the development of receptive grammatical abilities. Closer inspection of the individual data indicated that this occurs around the age of 10 years. This finding is in accordance with other investigations that have also reported diminished or ceasing progress in this domain around the age of 10–12 years (Chapman et al., 2002; Laws and Gunn, 2004; Conners et al., 2018). In contrast to the results by Chapman et al. (2002) and Laws and Gunn (2004), we did not find declining performance in grammar comprehension in our sample. However, in these studies decline was mostly observable in late adolescence and early adulthood, setting off around the age of 17 years. As the participants in the present study were younger than 17 years at study start (one exception) it remains open if they will possibly be affected by decline when they get older.

In the expressive domain, a large part of the investigated sample ($n = 12$) already performed well in verbal agreement marking at T1 and therefore only had limited potential for further development in this area. The descriptive analysis of the remaining five participants' results at T2 and T3 revealed consistent improvement only for two individuals. Both achieved a good proficiency with verbal agreement marking (accuracy score of over 80%) at some point between T1, when they were younger than 10 years, and T2 when they were slightly older than 10 years (chronological age: 11 and 13 years). In contrast, for three participants no consistent progress could be determined. They had not succeeded in mastering the system of verbal agreement marking by T3 when they were 15–20 years old. Thus, taking into account the small number of participants one can cautiously conclude that individuals who have not reached a high level of proficiency with respect to subject-verb agreement marking by late childhood might not display further development in this grammatical domain and, hence, do not acquire the German system of subject-verb agreement marking.

For the production of *wh*-questions, we saw high performance at T1 in five participants. The others exhibited a significant positive change when T1 and T2 performance were compared at group level. Difference scores were not related to the chronological age of the participants, suggesting that, in contrast to the receptive domain, improvements in *wh*-question production occurred irrespective of age. However, a closer inspection of the present data indicates that a notable improvement in the production of *wh*-questions was only observable for those participants who had acquired verbal agreement marking. Reversely, little change could be seen in those participants that had not acquired verbal agreement marking until late childhood (i.e., around the age of 10 years). The data, thus, suggest an implicational relationship between the acquisition of the verbal agreement system and progress in the production of *wh*-questions: progress in *wh*-question production could only be observed in those individuals who had mastered the system of verbal agreement marking.

The implicational relationship between these two phenomena is reminiscent of morphosyntactic development in typically-developing two-to-four-year old German-speaking children where the mastery of the verbal agreement system also precedes the production of *wh*-questions (Clahsen and Penke, 1992; Penke, 2001). The developmental relation between verbal agreement inflection and the production of *wh*-questions is rooted in the V2-property of German: in main clauses the finite verb, i.e., the verb that is marked for subject-verb agreement, moves to the second structural position in the clause. In the framework of generative syntax, this is achieved by movement of the verb from a position within the verbal phrase (VP), which encodes the argument structure of the described event, to functional projections which serve syntactic functions. An inflectional phrase (IP) takes care of the agreement inflection between subject and verb; another functional phrase (CP) accounts for the appearance of the finite verb in the second structural position in the sentence. To appear in this syntactic position the verb undergoes two syntactic movement operations: It first moves from the head of the VP to the head of the IP to enter into an agreement relation with the subject. Subsequently, it moves from the head of the IP to the head of the CP to appear in the second structural position (Haegeman, 2001).

An exemplification is presented in Figure 4 which displays the syntactic tree associated with a short German *wh*-question such as *Wen kitzelt der Junge?* ("Who is the boy tickling?"). In the VP, the verb describing the action (*kitzeln* "tickle") occupies the head position, the Agent of the action ("the boy") is situated in the specifier position of the VP (SpecVP) and the Theme/Patient of the action is lexicalized by a *wh*-pronoun in the complement position of the verbal head. In a first round of syntactic operations, the Agent moves to the specifier position of the IP where it is marked as subject by receiving nominative case inflection. The verb moves to the head of the IP to enter into an agreement relation with the person and number specifications of the subject, expressed by subject-verb agreement markers on the verb. Each moved constituent leaves behind an indexed trace (*t*) that connects the moved constituent to its base position in the VP. In the next round of syntactic operations, the finite verb moves from the head of the IP to the head of the CP position to occupy the second structural position in the sentence. V2 word order then comes about by movement of another sentence constituent, here the *wh*-pronoun, to the specifier position of the CP, the sentence initial position (so-called *wh-movement*).

In the lexical learning framework proposed by Clahsen et al. (1996), the acquisition of the verbal agreement system that takes place between the ages of two-to-three in typically-developing German-speaking children brings about the build-up of both functional phrases, the IP and the CP in the syntactic tree, thus, enabling the V2 movement of finite German verbs. The acquisition of the verbal agreement system leads to the build-up of the IP in the syntactic tree. With the acquisition of subject-verb agreement, the child can now identify which verbs move to the second structural position in main clauses, the head of CP, i.e. verbs inflected for subject-verb agreement. Moreover, s/he has acquired the means to inflect all verbs for agreement with the subject. Besides the head position for the finite verb, the CP contains a specifier position (SpecCP) that can now be filled with a *wh*-phrase moved out of its position in the VP. Movement of the *wh*-phrase to sentence-initial position can, thus, only occur after the build-up of the CP layer (by age 3 to 4 years in typically-developing German-speaking children) that is itself connected to the acquisition of subject-verb agreement marking. This is in line with the results of the implicational analysis reported above which showed that mastery of the verbal agreement system consistently preceded the ability to produce syntactically correct *wh*-questions in our participants. This observation suggests that the acquisition of the system of verbal agreement marking might have a trigger function for further grammatical development in individuals with Down syndrome. Moreover, our data suggest that the building of the syntactic tree needs to be completed within a certain time window, around the age of 10 years (chronological age), to enable further grammatical development with respect to syntactic structures that require the projection of the CP layer, such as *wh*-questions. While this is an intriguing suggestion, its data base is small and requires further investigation.

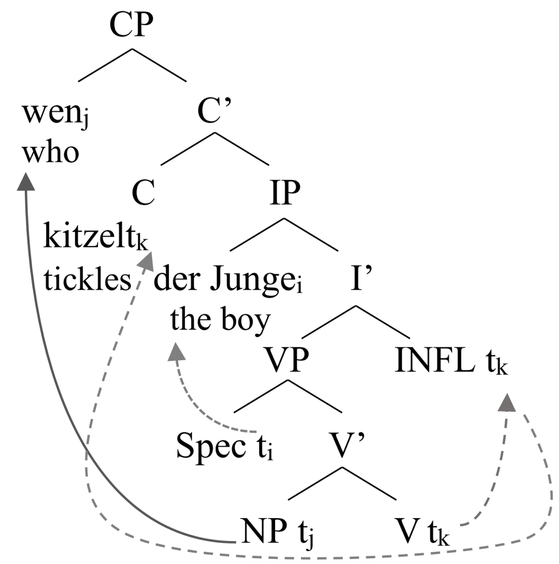

FIGURE 4
Syntactic tree of the *wh*-question *Wen kitzelt der Junge?* ("Who is the boy tickling?").

Note that while the acquisition of the system of subject-verb agreement inflection plays a central role for the acquisition of syntactic structures in German, this does not hold across languages. The lexical learning approach to syntactic development assumes that the acquisition of functional heads proceeds when children acquire the bound or free grammatical morphemes that lexicalize these functional heads in the language they acquire (Clahsen et al., 1996). While this acquisition procedure holds across languages, the lexical elements that lead to the build-up of functional phrases are language-specific. Thus, while the implicational relationship between the acquisition of subject-verb-agreement and the build-up of the CP layer holds for German, future research would have to target whether similar implicational relationships characterize the acquisition of syntactic structures in other languages and whether developmental restrictions in the timely build-up of the syntactic tree can also be observed in individuals with Down syndrome that speak other languages than German.

Our suggestion that there is a critical window for the acquisition of verbal agreement inflection and, concomitant, the building of the syntactic tree, and that only if this is accomplished, further grammatical development might come about in German-speaking individuals with Down syndrome conforms to the findings of Conners et al. (2018). In their longitudinal study, they observed no progress with respect to expressive grammatical abilities in the tested individuals with Down syndrome. Interestingly, their participants were aged 10 to 21 years at T1, suggesting that a critical time window for further syntactic development (around the chronological age of 10 years) might already have been closed for their participants. This supports our findings and the suggestion that there is a critical window for the development of expressive grammatical abilities in individuals with Down syndrome that might close around late childhood.

Taken together, our data indicate a critical time window for the development of receptive as well as expressive grammatical abilities in individuals with Down syndrome that seems to close around late childhood. Within this critical time window there is the possibility of further progress in receptive and expressive grammatical abilities. However, this does not imply that progress within this critical time window is guaranteed to occur. In the present study, for instance, there was one participant (P4) who displayed no consistent progress in receptive and expressive grammar measures despite an age of only 7 years at T1. More research is needed to confirm the conclusions drawn from the present data and to determine the factors that advance or hinder further development of grammatical abilities within the critical time window in individuals with Down syndrome.

4.2. Relation between morphosyntactic development and nonverbal cognition as well as verbal short-term memory (RQ2)

A further goal of our study was to investigate whether individual grammatical development is influenced by nonverbal cognitive abilities or verbal short-term memory capacity, two factors that have been discussed as predictors of grammatical development in the literature.

With respect to progress in nonverbal cognition, we found ongoing development in most of the examined individuals irrespective of their chronological age. The finding of continuing growth of nonverbal cognitive abilities in adolescence in individuals with Down syndrome is consistent with existing studies (Couzens et al., 2011; Channell et al., 2014). It contrasts, however, with the stagnation of receptive and expressive grammatical abilities that we observed in participants after late childhood. Correlational analyses between reasoning scale raw scores at T1 and difference scores for our grammar measures did, therefore, not reveal a relation. Specifically, it was not the case that participants with higher scores in our measure of nonverbal cognition at study start showed more improvement in either receptive or expressive grammatical abilities than those with lower scores, a relation that one might have expected given the findings of previous cross-sectional research that found positive correlations between nonverbal cognitive and language performance (Chapman et al., 1991; Abbeduto et al., 2003; Aktaş, 2004; Price et al., 2007, 2008; Iacono et al., 2010; Estigarribia et al., 2012; Finestack et al., 2013). Our finding that progress in nonverbal cognition is not related to progress in grammar development is, however, in accordance with Chapman et al. (2002) who also found that their measure of nonverbal cognition did not add to the predictive power in their models for syntax comprehension and production. Although our data base is limited, especially with respect to the relation between expressive grammatical abilities and nonverbal cognition, the data presented here and the data of the other longitudinal study investigating nonverbal cognitive and language development (i.e., Chapman et al., 2002) provide converging evidence that development in nonverbal cognitive abilities in children and adolescents with Down syndrome does not proceed hand in hand with ongoing development in grammatical abilities. This does not preclude the possibility that such a relation might hold for very young children with Down syndrome, an issue that should be targeted by further research.

With respect to the development of verbal short-term memory capacity, measured via nonword repetition, the present study indicated that growth in this domain also levelled off early (around the chronological age of 10 years). There was no significant difference in nonword repetition performance between T1 and T2 at group level. Moreover, performance in nonword repetition displayed a negative correlation with chronological age, indicating less growth or even a decline in nonword repetition performance in older compared to younger participants. Notable individual improvement did not occur after the age of 10 years and decline was observable in three individuals, two of them being the two oldest. An early termination in the development of verbal short-term memory capacity has also been reported by Conners et al. (2018) and Laws and Gunn (2004). In their studies, nonword repetition performance even declined in most participants with a chronological age over 10 years. Concerning the role of verbal short-term memory for grammatical development, the correlational analyses provided no indication that participants with better performance in the verbal short-term memory task at study onset exhibited larger growth in receptive or expressive grammatical abilities. The finding for the expressive domain conforms to the analyses by Chapman et al. (2002), where verbal short-term memory also did not prove to be a predictor for growth in grammar production. With respect to receptive grammar, a significant relationship to verbal short-term memory as found in other longitudinal studies (Chapman et al., 2002; Laws and Gunn, 2004) is not confirmed by the present results. Note, however, that the correlation between T1 nonword repetition scores and T2 performance in receptive grammar in Laws and Gunn's (2004) investigation was only evident in a subsample, aged below 14;8 years at T2. This younger subsample also showed more consistent progress in grammar comprehension than the older participants. Taken together, there is no indication that verbal short-term memory performance is the decisive factor for grammatical development in individuals with Down syndrome.

4.3. Other potential predictive factors

Apart from chronological age, nonverbal cognitive abilities or verbal short-term memory capacity, there are others factors that could potentially be related to language progress in individuals with Down syndrome. One factor, that comes to mind, is ongoing support in the form of speech and language therapy. However, a subsequent inspection of this aspect in our participants did not reveal a relation between improvements in language abilities or the lack thereof and the application of language intervention. Of the ten participants that showed little or no change in receptive grammatical abilities between T1 and T2 five had received speech and language therapy during the entire time between T1 and T2. The other half did not obtain language intervention or, in one case, only for a limited part of the time. Furthermore, all five individuals that displayed ongoing difficulties with the production of *wh*-questions received language intervention throughout the duration of the study. Thus, it rather seems to be the case that more severe limitations in language abilities, especially in the expressive domain, are met with prolonged therapeutic services.

Another factor that might be beneficial for language development is the acquisition of literacy. Information on the participants' reading abilities was only collected at T2. Therefore, it cannot be determined whether the degree of literacy had any influence on the grammatical development of the investigated individuals between T1 and T2. However, at T2 only one participant could not read and one could only read short, frequently occurring words. Three participants had reading skills at sentence level. The majority of the sample (12 out of 17) was able to read at text level. That included participants who did not show substantial progress in receptive or expressive grammatical abilities which suggests that for them literacy did not advance grammatical development.

In addition, we did not find a relation between parents' level of education, measured on a 9 level scale (ranging from 0 = early childhood education to 8 = doctoral degree) according to the International Standard Classification of Education (United Nations Educational, Scientific and Cultural Organization, 2012), and language change (Spearman's correlations between mother's and father's level of education and difference scores for receptive grammar and *wh*-production: $p > 0.5$ each).

To summarize, the present data, even though limited, do not provide evidence that factors other than chronological age, such as speech and language therapy, literacy or parents' level of education, determine which participants show progress and which exhibit little or no improvement.

4.4. Limitations

There are some limitations of the current study we would like to address. The first issue concerns the relatively small sample size of 17 individuals with Down syndrome—and an even smaller sample in the analyses of expressive grammar—in our study that limits statistical analyses. Although, we started with a relatively large sample size of 31 participants at T1, a large number of these could unfortunately not be recruited again for subsequent testing at T2. In addition, the age range was quite large. Furthermore, the time intervals between T1 and T2 varied between 4½ and 6½ years. Despite these limitations, core findings of our results—such as the levelling off in the development of receptive grammar—concur with previous studies that tested larger samples (e.g., Conners et al., 2018). Thus, a consistent picture of the development of expressive and receptive grammatical skills emerges, pointing to a critical developmental window of about 10 years of age. In future studies, however, more testing points with equal time intervals before and after the age of 10 years should be scheduled, to determine the time window for the acquisition of specific grammatical skills in individuals with Down syndrome more exactly.

Another issue concerns the composition of the sample. Due to the inclusion criteria, such as monolingualism and verbal means of communication, the sample might not be truly representative of the population of individuals with Down syndrome. Furthermore, other background data such as information on ethnicity or adaptive functioning was not available and should be gathered in future studies. Likewise, more detailed information on the methods, goals, and intervals of past therapeutic interventions would be desirable to explore which factors limit or boost an individual's potential for grammatical development.

Regarding experimental procedures, contrary to other studies (e.g., Chapman et al., 2002), we did not use MLU as a global measure for expressive grammar but focused on specific morphosyntactic phenomena. This limited the comparability of our results to previous findings regarding the development of expressive grammatical abilities. Also, given that only a limited set of morphosyntactic phenomena can be tested without overtaxing the participants, an advance selection had to be made. Thus, we might have missed aspects of morphosyntax that exhibit different developmental patterns in individuals with Down syndrome. Another limitation is the use of two different, albeit very similar, tasks to assess the production of *wh*-questions at T2. Also, information on reliability and validity is not available for the experimental tasks that were used to test expressive grammar.

The SON-R 2.5-7 was used to assess nonverbal cognition. The limited age band of the SON-R norming sample did not allow to calculate IQ scores for most participants to provide them as background information. Therefore, we reported nonverbal mental age equivalents to describe the sample. A ceiling effect in this regard was evident for two participants at T2. Thus, only their minimum mental age could be determined. Note, that nonverbal mental age equivalents have several limitations (see Maloney and Larrivee, 2007 for a comprehensive examination of age equivalent scores).

Ability scores such as growth scale values, that are weighted for item difficulty and measured on an interval scale, are not available for the standardized tests that were used in this study. Hence, we relied on raw scores which do not follow an equal-interval scale. Statistical measures that would indicate significant individual changes were not applicable to our data. Therefore, we—somewhat arbitrarily—considered an increase of three or more raw points as a notable increase in TROG-D or nonword repetition performance. However, as indicated by the data (in the figures and the supplementary table), a different setting for this value (to 2, 4, or even more points) would not change our main finding that larger increases only occur in younger participants.

4.5. Clinical implications

The results of the present study not only contribute to the understanding of possible developmental patterns in grammar development, but also lead to clinical implications. The focus of speech and language therapy in individuals with Down syndrome should always be based on the respective individual strengths and weaknesses, as identified by comprehensive diagnostic assessments. Nevertheless, some general conclusions can be drawn from the current findings.

The indication of a critical time window for the development of both receptive and expressive grammatical abilities, which might end around the age of 10 years, suggests that this period is particularly important for intervention in the area of grammar. This is highly relevant because, in our experience, supporting improvement in vocabulary or pronunciation is often prioritized over grammar in therapeutic settings, especially in childhood.

Previous research has shown various impairments in language comprehension in individuals with Down syndrome [*cf.* review by Andreou and Chartomatsidou, 2020]. We would therefore like to stress the importance of addressing not only expressive but also receptive abilities in language therapy, especially since limitations in language comprehension can easily be overlooked or remain unrecognized without suitable diagnostic instruments. Given the concurrent findings of a developmental window closing in late childhood, it is necessary to sensitize educational and therapeutic professionals for receptive deficits and the need for early detailed assessment and intervention in this respect. Therapeutic work on receptive grammar might also have a positive impact on grammar production. For example, Chapman et al. (2000) identified receptive language skills of people with Down syndrome as a crucial predictor for their productive abilities (MLU and number of different words in narration). Future intervention studies should therefore evaluate whether transfer effects from the receptive to the expressive language domain are indeed possible.

Regarding expressive grammatical development, based on the findings of our study, subject-verb agreement can be hypothesized to be a critical structure for the development of the syntactic tree and thus, for further expressive syntactic development in German. If this is indeed the case and if there is a critical time window for the acquisition of verbal agreement, a stronger emphasis on supporting its acquisition within that timeframe could possibly pave the way for further grammatical development in German-speaking individuals with Down syndrome – an issue that probably deserves longitudinal research. There are several therapeutic approaches to support the acquisition of verbal agreement inflection in German (e.g., training of final consonants (TraFiK); Penke et al., 2020; the psycholinguistic approach (PLAN); Siegmüller and Kauschke, 2006). Their effectiveness for individuals with Down syndrome should be investigated in future evaluation studies. Furthermore, to make the best possible use of the described time window, it might be advisable to administer not only regular outpatient speech and language therapy, but also treatment in the form of intensive therapy.

The emphasis we put on targeting grammar in language intervention in early ages to use important time windows for development does, however, not imply that progress in receptive or expressive grammatical abilities of individuals with Down syndrome is impossible beyond the age of 10 years or that therapy should be interrupted or terminated at this age. Progress in expressive grammar seems possible as soon as the syntactic tree is complete. Moreover, targeted speech and language therapy might in fact be necessary to avert stagnation or decline in language abilities, the more so since school support ends after the transition from adolescence to adulthood.

5. Conclusion

The findings of the present study indicate that development in receptive grammar levels off in late childhood, around the age of 10 years, confirming previous research. For expressive grammar, mastery of the verbal agreement system consistently preceded the ability to produce syntactically correct *wh*-questions in our participants. Notable improvement in *wh*-question production only occurred in individuals with good performance in subject-verb agreement marking. Thus, our study not only confirmed previous results but expanded them by putting forward the suggestion of a trigger function of the acquisition of the verbal agreement system for further expressive grammatical development in German-speaking individuals with Down syndrome. We hypothesized that the building of the syntactic tree, that is connected to the acquisition of the verbal agreement paradigm, needs to be completed within a certain time window, around the age of 10 years, to enable the acquisition of sentence structures that involve the CP layer, e.g., *wh*-questions. These ideas provide an avenue for future research and should be pursued in studies with larger longitudinal samples and different measures for expressive morphosyntactic abilities.

Our finding of a critical time window for further morphosyntactic development in individuals with Down syndrome has implications for speech and language intervention. Whether targeted intervention in adolescence can help to delay or even prevent the levelling off in grammatical development observed in individuals with Down syndrome, is an important issue to address in future research.

Data availability statement

The datasets presented in this article are not readily available because they include data from vulnerable participants and third-party availability of the data was not part of the ethics approval. Requests to access the datasets should be directed to BW: bwitecy@uni-koeln.de.

Ethics statement

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

Author contributions

MP conceptualized and supervised the study. BW, EW, and IN collected the data. BW and MP analyzed the data. BW wrote the manuscript. EW, IN, and MP contributed to the writing of the manuscript. BW, EW, IN, and MP edited the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2023.1118659/full#supplementary-material

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A biophysiological framework exploring factors affecting speech and swallowing in clinical populations: focus on individuals with Down syndrome

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Speech and swallowing are complex sensorimotor behaviors accomplished using shared vocal tract anatomy. Efficient swallowing and accurate speech require a coordinated interplay between multiple streams of sensory feedback and skilled motor behaviors. Due to the shared anatomy, speech and swallowing are often both impacted in individuals with various neurogenic and developmental diseases, disorders, or injuries. In this review paper, we present an integrated biophysiological framework for modeling how sensory and motor changes alter functional oropharyngeal behaviors of speech and swallowing, as well as the potential downstream effects to the related areas of language and literacy. We discuss this framework with specific reference to individuals with Down syndrome (DS). Individuals with DS experience known craniofacial anomalies that impact their oropharyngeal somatosensation and skilled motor output for functional oral-pharyngeal activities such as speech and swallowing. Given the increased risk of dysphagia and "silent" aspiration in individuals with DS, it is likely somatosensory deficits are present as well. The purpose of this paper is to review the functional impact of structural and sensory alterations on skilled orofacial behaviors in DS as well as related skills in language and literacy development. We briefly discuss how the basis of this framework can be used to direct future research studies in swallowing, speech, and language and be applied to other clinical populations.

KEYWORDS

speech, swallowing, sensorimotor control, Down syndrome, biophysiological framework

Introduction

This article proposes a multidimensional theoretical framework for understanding how characteristics associated with the phenotype of Down syndrome (DS) may influence performance of swallow behavior and production of intelligible speech, as well as impacting language development and foundational literacy outcomes such as phonemic awareness and phoneme-grapheme correspondence. This framework takes as its starting point a model introduced by the first and third authors that considered food selection, swallow, and speech in healthy older individuals (Etter and Madhavan, 2020). This model was developed because of the unique needs in healthy older adults. One example is that older adults without a diagnosis related to dysphagia typically do not report swallowing changes, and instead make self-determined compensations like changing their diet or avoiding foods, which may be the result of early dysphagia and lead to negative consequences (Roy et al., 2007; Madhavan, 2020). These needs are often missed in models that are applicable to clinical populations.

We chose to expand this integrated framework to the DS population because despite our knowledge of heightened swallowing, speech, language, and literacy problems in individuals with DS, it has not consistently translated to improved clinical outcomes. This may be because management approaches used in DS often "borrow" techniques from other populations (Neil and Jones, 2018), however, these other populations do not have the syndrome-specific structural, functional, or physiologic dysmorphologies characteristic in DS. To improve precision interventions in DS, an integrated understanding of the unique phenotypical characteristics is an important early step. The framework is also consistent with the World Health Organization's International Classification of Functioning (Centers for Disease Control [CDC], n.d.), with a particular focus on the effects of the characteristics associated with DS on body functions and structures and, in turn, activities and participation. Although we acknowledge the critical role of environmental factors as well, for the sake of space we focus primarily on the body functions and structures within DS. Moreover, our approach is consistent with recommendations for critical directions in DS research as suggested by an expert panel (Hendrix et al., 2020). Our extensions in DS include: (1) application of the framework within the context of the distinctive phenotypically linked structural and functional characteristics associated with DS, and (2) consideration of the potential downstream impacts of the phenotypic oropharyngeal characteristics in DS on language and early literacy skills, thus extending the analysis beyond swallowing to speech and related linguistic functions. **Figure 1** presents the framework, with the original conceptualization along with the extension to language and literacy, as it relates to DS.

Phenotypic characteristics in DS of relevance for the framework

In the United States, it is estimated that approximately 8 per 10,000 individuals are living with DS, a genetic disorder that results from a full or partial extra copy of chromosome 21 (Presson et al., 2013). Difficulties with speech intelligibility, swallowing, and language and literacy are reported throughout the lifespan in DS and are likely in part due to phenotypically linked structural, linguistic, and cognitive characteristics (Bruni et al., 2010). For instance, phenotypical differences in the structure and function of oral-motor mechanisms and in measures of cognition and language are well-documented in individuals with DS. An example of a structural change includes craniofacial anomalies that may cause obstruction in the airway at multiple levels in the respiratory system (Shott and Donnelly, 2004). These may impact functional speech production by affecting the motor processes involved in speech kinematics, in turn affecting speech intelligibility.

In addition to impacts on swallow and speech, the phenotypic characteristics in DS likely also impact both language and literacy outcomes across the lifespan. As we will outline in the second half of the paper, functions of language and literacy may also be affected both by difficulties in producing intelligible speech as well as potentially in hearing or perceiving spoken input. Although language and literacy learning are often considered to occur primarily in early to middle childhood, in reality these are lifelong learning activities, particularly for individuals with DS. For instance, Chapman et al. (2002) demonstrated through growth curve modeling that although the speed of growth changes, there is continued growth in both expressive and receptive syntax throughout adolescence in DS (Chapman et al., 2002). As Abbeduto and Thurman (2022), p. 1583 point out, literacy instruction often receives less attention for "students with intellectual disabilities as they get closer to exiting formal schooling and transitioning to adulthood, despite the reality that independence in adulthood depends critically on language and literacy." However, the new motivations introduced by access to social media and for vocational skills in adolescence and adulthood mean that literacy too should continue to evolve across the lifespan, a point raised also raised by Abbeduto et al. (2007). Adding a further layer of complexity, individuals with DS demonstrate accelerated aging, especially in the brain (Lott and Head, 2005). Accelerated aging in the brain is thought to be as significant as 11 years earlier (Horvath et al., 2015), indicating that even though learning of academic skills continues into the third decade, individuals with DS also start to experience loss of skills much earlier than the general population. In the next sections, we briefly review the evidence under each of the primary factors in our proposed framework as related to swallow function, speech production, and language/literacy in individuals with DS.

Structural: oral-pharyngeal and neuroanatomical differences in individuals with DS

Normal swallowing depends on the rapid transfer of the prepared food or liquid bolus from the oral cavity to the stomach. To achieve manipulation, mastication, and containment in the oral cavity, coordinated movement between the lips, tongue, and jaw are crucial. As the bolus is transferred from the oral cavity posteriorly through the aerodigestive tract, adequate functioning of the soft palate, the larynx, and close coordination with the respiratory mechanism are important. Oropharyngeal anatomy in DS has distinctive features, even with expected intra-individual variations. These distinctive features are particularly relevant in the structures involved in swallowing. Common facial features include reduced mouth width and prominent lips, reduced size of hard palate, variety of dental anomalies, and relative macroglossia (Sforza et al., 2012). Compared to those without DS, individuals with DS have mid-face hypoplasia (Uong et al., 2001), a relatively small maxilla, but typical sized mandible (Allanson et al., 1993), dysmorphology of cranial base, maxilla, and mandible (Suri et al., 2010), and reduced palatal volume (Bhagyalakshmi et al., 2007; Dellavia et al., 2007). Additionally, airway abnormalities such as laryngomalacia, tracheomalacia, and bronchomalacia are frequent in individuals with DS (Bertrand et al., 2003).

Neuroanatomical differences may also play a role in the functioning of these complex tasks. Recent neuroanatomical studies, including MRI studies of adolescents with DS, have reported smaller cerebellar volumes, compared to age matched neurotypical peers, and other structural brain differences that are

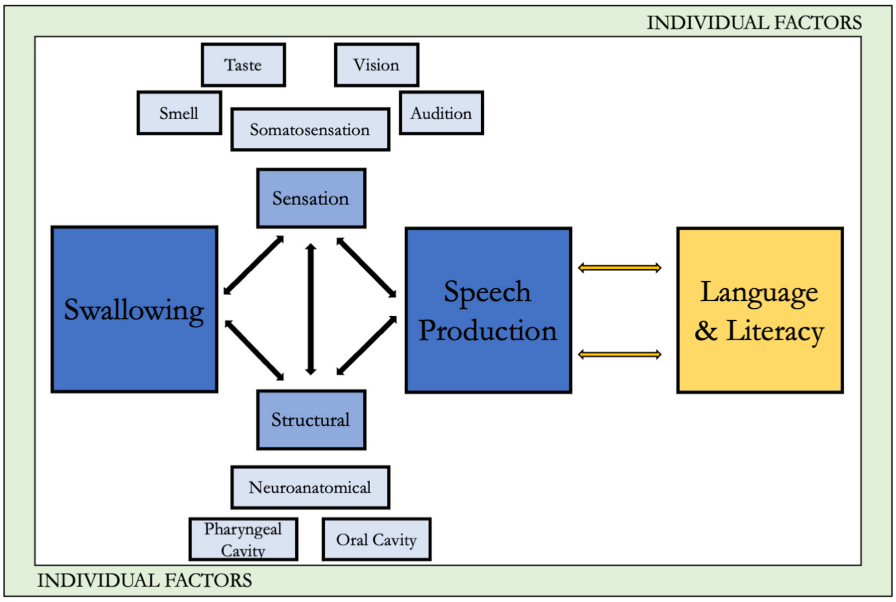

FIGURE 1
A biophysiologically-integrated framework of swallow, speech, and language/literacy.

relevant to swallowing, speech, and language disorders (Wilson et al., 2019). Evidence suggests reduced volumes of total gray and white matter, cortical lobar, hippocampal, and cerebellar regions (Hamner et al., 2018). Individuals with DS may experience premature brain aging, with accelerated volume loss. The incidence of age-related cognitive decline and dementia is greater in adults with DS compared to the general population and develops earlier in life (Cole et al., 2017). In fact, older patients with DS show neuropathological changes characteristic of Alzheimer's Dementia, including increased cerebral beta-amyloid deposits, neurofibrillary tau tangles, neuritis plaques, and neuron cell loss (Cole et al., 2017). Collectively, these anatomical changes could impact swallowing, speech, and language outcomes in individuals with DS.

Sensation: sensory differences in individuals with DS

Sensory information allows individuals of all ages to internally perceive, recognize, and engage with their external environment (Bruni et al., 2010). Because each movement has a sensory consequence, traditional motor control theories highlight the tight temporal synchrony between sensory information and motor response needed for learning and maintaining skilled behaviors (Shumway-Cook and Woollacott, 2012). Thus, any alterations in sensation may impact motor plans and any alterations in structures could influence sensory feedback. One motor control theory that applies this concept for learning speech is the Direction into Velocity of Articulators (DIVA) model (Tourville and Guenther, 2011). The purpose of this computer-generated model of speech development is to highlight how children learn speech motor control through the interaction between auditory and somatosensory feedback from motor movements. Briefly, each time a child babbles, they receive auditory and somatosensory feedback that can be used to inform their motor plan. Each successive babble or speech attempt provides more information for the child to update their motor plan. As children learn, they continually update their motor plans through sensory feedback. Using the DIVA model as a basis, it is clear that oropharyngeal somatosensation, along with audition, is a crucial element for learning accurate speech production (Guenther et al., 2006; Golfinopoulos et al., 2010).

Hearing

Approximately 38–78% of individuals with DS experience hearing loss (Intrapiromkul et al., 2012; Clark et al., 2017). Changes in auditory sensation can be conductive or sensorineural, but both may be linked to structural alterations. In several studies, conductive hearing loss was found to occur in 1/3rd of the study participants with DS, and typically secondary to chronic ear infection and stenosis of the external auditory canal (Park et al., 2012; Clark et al., 2017). Sensorineural hearing loss in this population is also seen, with computed tomography scans showing inner near abnormalities including stenosis of the cochlear nerve canal and internal auditory canal in 25% individuals with DS (Intrapiromkul et al., 2012; Nightengale et al., 2017). As we will describe, hearing loss may contribute to difficulties with speech perception and phonological processing, thus also contributing to difficulties in development of oral language and emergent literacy skills (Laws and Hall, 2014; Manickam et al., 2016).

Vision

Visual acuity development in DS has been found to follow a different developmental trend than typical peers (Purpura et al., 2019). Structures with reported abnormalities include the lid, iris, lens, retina, and cornea (Krinsky-McHale et al., 2014). These structural changes result in the increased prevalence of nystagmus, strabismus, astigmatism, and significant refractive errors in individuals with DS (Krinsky-McHale et al., 2014). Across the lifespan, the incidence of visual impairments increases with age such that by 60 years of age, 85% of individuals with DS had visual impairment (Krinsky-McHale et al., 2014). The sensation of vision has a limited role in a biophysiological model of speech and swallowing; however, vision deficits can have implications for learning, particularly literacy learning, cognitive functioning, and adaptive behavior. Additionally, vision can play a role in priming a person for efficient swallowing, specifically through its role of stimulating saliva production needed for the oral preparatory phase of swallowing.

Somatosensation

The somatosensory system transmits touch, pressure, and relative body position information from peripheral receptors centrally to the brain to inform movement responses. This system may be impaired in individuals with DS (de Knegt et al., 2015). Using quantitative sensory testing methods, de Knegt et al. (2015) assessed 188 adults with DS to determine their abilities to discriminate temperatures, sharp and dull pressure, and to detect touch on their forearm. A decreased ability to distinguish between sharp-dull pain was found to be associated with IQ level as measured on a standardized test. Lower sensitivity to pain may be the result of a smaller mediodorsal thalamic nucleus in DS, as this structure is important in transmitting sensory information to the prefrontal cortex (de Knegt et al., 2015). The loss of high-quality somatosensory feedback can interfere with the ability to learn and maintain accurate motor plans necessary for speech and swallowing. It is possible some deficits in behavior may not be the result of peripheral sensory appreciation, but in the processing and use of sensory inputs for accurate motor planning (Will et al., 2019).

Sensory processing

Sensory processing is the continuous integration of information from the senses, movement, and muscle position by the nervous system which monitors an individual's response, including over- or under-responsiveness, difficulties with stimuli discrimination, and challenges with proprioception and motor planning (Miller et al., 2007; Will et al., 2019). Difficulty with sensory processing is common across individuals with neurodevelopmental disabilities and linked to maladaptive outcomes (Baranek et al., 2018). Using Dunn Sensory profiles, (Dunn, 1999) sensory processing and visual organization abilities of 206 children with DS were studied by Wuang and Su (2011). About 41% of their sample was reported to have a "definite difference" in low registration, 40% a "definite difference" in low endurance/muscle tone, and 39% showed a "definite difference" in sensory sensitivity (Wuang and Su, 2011). In a second study, almost half of children with DS experienced a definite difference in the low energy/weak, under-responsive/seeks sensation, and the auditory filtering domains (Bruni et al., 2010). The combined results of these studies point to differences in the way children with DS identify, process, and respond to various types of sensory information. These issues that arise in childhood might be expected not just to continue, but potentially to be magnified across the lifespan (Grieco et al., 2015).

Individual and environmental factors

In addition to the above factors, several individual and environmental factors can impact functional behaviors. Some of these individual factors include cognition (Anil et al., 2019), dietary requirements for nutrition and medical needs (Wallace, 2007), food preferences (Field et al., 2003; Anil et al., 2019) etc. Although intellectual disability is a characteristic of DS (and thus could be considered within the "DS phenotype" as well), we have chosen to consider it an "individual" factor instead. In part, this reflects the broad spectrum of intellectual and adaptive functioning found in individuals with DS (Mégarbané et al., 2013; Carr, 1988). Additionally, a thorough evaluation of the impact of some of the characteristic cognitive features (related to attention, memory, etc.) would require a dedicated article of its own, beyond the scope of the current article.

Environmental factors such as parental anxiety and grief surrounding a DS diagnosis (Carr, 1988), cultural expectations, and access to care can impact speech, swallowing, and literacy outcomes (McCabe et al., 2011; McGrath et al., 2011; van den Driessen Mareeuw et al., 2020). Although discussing all these factors is beyond the scope of this paper, in the following section we include a discussion on the importance of cognition on speech, swallowing, language and literacy.

Functional implications for swallow, speech, and language/literacy outcomes

In this section, we explore the potential interrelations between the information reviewed in the previous section and the functional outcomes of swallow, speech, language, and literacy. We first begin by considering the potential relationships at a broad/general level. We then offer two detailed examples of how phenotypic characteristics of DS could specifically affect each of the four functional outcomes of interest, as a model for how the other information in the upcoming section might play out across the functional domains. Not all of the direct relationships have been studied and warrant direct research.

Functional implications for swallowing

While seemingly effortless for most adults, the production of a safe swallow and intelligible speech involves rapid and complex coordination of oral-motor structures and functions. This coordination: (a) relies on high-quality sensory feedback from the lips, tongue, jaw, and pharynx; (b) requires skilled, coordinated motoric control, and (c) is informed by and dependent on cognitive, linguistic, and perceptual skills for planning and execution.

Extensive research has documented significant problems with swallowing in both children and in adults with DS. For example, dysphagia has a higher documented prevalence in adults with DS relative to controls (Capone et al., 2020; Chicoine et al., 2021), and adults with DS are substantially more likely to die from choking than those without DS (Landes et al., 2020). Feeding/swallowing difficulties are likely common in individuals with genetic conditions due to the complex interaction between medical, anatomical, physiological, and behavioral factors. In a study by Anil et al. (2019), the parents of 17 children with DS and 47 typically developing children completed a questionnaire regarding feeding. The most prevalent feeding problems in the oral phase were increased oral hold, increased duration for bolus manipulation, difficulty chewing, and inappropriate oral transit (Anil et al., 2019). In the pharyngeal phase, delayed posterior transit and aspiration were reported. In the esophageal phase, the researchers postulated that reduced muscle tone may result in increased vomiting, poor digestion, and gastroesophageal reflux (Anil et al., 2019). Additionally, considering sensory information that's important to swallowing, changes to taste and smell have been identified in individuals with DS across the lifespan, possibly related to structural differences that may impact nasal health, resulting in hypoplasia (Chen et al., 2006). Taste and smell deviations can impact swallowing because they are an important sensory input element for the motor output of an efficient swallow.

Oral-motor skills can also be impacted, with possible weak lip closure, compression pattern without the use of intraoral suction, tongue thrusting, and chewing difficulties (Cooper-Brown et al., 2008; Anil et al., 2019; Ross et al., 2019). In a retrospective chart review of 158 children with DS, oral motor difficulties occurred in 63.8% and oral sensory difficulties in 20.3% of the sample (Jackson et al., 2016). Oral sensory difficulties included both oral hyposensitivity and oral hypersensitivity. Ross et al. (2019) found that many children with DS only ate "easy" low-textured food and refused to chew. This downstream effect of oral sensory changes and textural preference can result in a lack of diversity in dietary and nutritional intake (Ross et al., 2019).

In the pharyngeal phase of swallowing, studies report frequent "silent" aspiration and what was characterized as deep laryngeal penetration in most or all participants (Frazier and Friedman, 1996; Jackson et al., 2016). Evidence suggests aspiration may be related to hypotonia of the pharyngeal musculature in infants, perhaps suggesting a generalized hypotonicity in individuals with DS (Shott, 2006). The lack of a cough response to aspirated materials indicates decreased laryngeal sensation.

In addition to dysphagia, individuals of all ages with DS can present with chronic pulmonary problems and obstructive sleep apnea, that contribute to respiratory problems like recurrent pneumonias, recurrent upper and lower respiratory tract infections, and even respiratory failure (Bertrand et al., 2003). Feeding and swallowing difficulties thus become more significant due to an increased risk of aspiration, lower immune system response, and possible support needs for activities of daily care and living. In fact, respiratory illness is one of the most common causes of mortality in DS (Landes et al., 2020). Aspiration from food and liquid ingestion is as a top cause for respiratory illness and mortality in children and adults with DS (Frazier and Friedman, 1996; Landes et al., 2020). Future studies should analyze whether comorbidities that are associated with DS are prognostic of the presence, severity, and longevity of dysphagia.

Finally, the act of swallowing requires coordination between physiologic and sensorimotor responses, visual recognition of food, motor planning, wish to eat, and essentially, cognitive awareness (Rogus-Pulia et al., 2015). Particularly in the oral phase of swallowing, cognitive deficits in attention, decision making, recognition and orientation can impair swallowing (Langmore et al., 2007). Because decreased attention and impulsivity are frequently reported in individuals with DS (Capone et al., 2006), they may be at risk for increased difficulties in certain aspects of swallowing, although this possibility requires direct study. Additionally, the brain volume loss and premature aging experienced by individuals with DS may lead to the need to adapt these deficits to a constantly evolving mechanism, potentially creating new and lifelong difficulties with complex oral motor behaviors.

Functional implications for speech, language, and literacy outcomes

The combination of structural anomalies, peripheral sensory changes, alterations in sensory processing and individual factors such as cognition likely impact speech behavior. Across the lifespan, individuals with DS experience difficulties with intelligible speech that impact vocational social, independent living, and self-advocacy outcomes, among others (Kumin, 1994; Fawcett and Peralego, 2009; Kent and Vorperian, 2013). Children developing typically usually reach 100% intelligible speech by 4 years of age however, it is unusual for children with DS to reach 100% speech intelligibility at that age (Kumin, 2006). Indeed, Martin et al. (2009) note that "nearly all individuals with DS may be difficult to understand at least some of the time" (p. 115). Hearing loss and auditory discrimination difficulties make it more difficult for children with DS to perceive the subtle differences between sounds, which again may contribute to the difficulty in producing speech sounds (Kumin, 2006) as well as learning foundational literacy skills such as phoneme-grapheme correspondence.

Physiologic findings suggest that speech and voice problems such as dysarthria, apraxia, voice and resonance problems may be associated with features such as limited tongue moment during vowel production which results in reduction in acoustic vowel space, articulatory working space, and articulatory speed (Wilson et al., 2019). Other factors that are associated with their speech and voice disorders include craniofacial and laryngeal dysmorphologies, motor impairments, phonological delay or disorder, dysfluency, and hearing loss (Rosin et al., 1988; Kent and Vorperian, 2013; Wong et al., 2015).

Little direct research has examined possible relations between speech production, cognition, and language and literacy outcomes. However, Cleland et al. (2009) evaluated whether global measures of language and cognitive functioning correlated with overall intelligibility in 15 youth with DS; they found little correlation. However, we would argue language-speech relationships are not global (as measured by Cleland et al., 2009), but rather represent more specific relationships between speech production and cognitive and linguistic demands (such as working memory and/or syntactic complexity, respectively). For instance, expressive grammar is a particular challenge in DS (Rvachew and Folden, 2018; Abbeduto and McFadd, 2021). Studies of children with typical development and those with language impairments have demonstrated a "trade-off" between speech and language, such that when linguistic demands increase, speech movement becomes more variable and phonemic accuracy decreases (Masterson and Kamhi, 1992; Maner et al., 2000; Seeff-Gabriel et al., 2010). Given this speech-language tradeoff in other populations, and the selective difficulty in grammar in DS, it is possible that when demands of either speech or of language increase, there is a toll on the other. An individual seeking to produce a particularly difficult spoken token ("crocodile") may sacrifice syntactic complexity, producing it either in the context of less-complex syntax and/or making syntactic errors.

Sensation limitations also have implications for language and/or literacy outcomes. Reduced access to the speech signal likely affects speech perception as well as speech production. Difficulties with speech perception or processing in turn affects language comprehension, at least for spoken input. For instance, a child who cannot distinguish between minimal phonetic pairs ("bat"/"pat") may in turn have difficulty producing them correctly in their speech, and with mapping the words to their respective semantic concepts linguistically and with acquiring the phoneme/grapheme linkages needed for literacy (e.g., see Abbeduto et al., 2007, for a discussion of the role of auditory and phonological processing on language and literacy outcomes in individuals with Down syndrome). A child who is not perceiving final sounds, such as/t/or/d/, will in turn be challenged in incorporating those into expressive or receptive syntax, as many morphemes occupy that final position. Limitations in vision will compound the difficulties with phoneme-grapheme acquisition as well as other literacy outcomes (whole word reading, decoding; Woodhouse, 2005). Finally, limitations in oral somatosensation can result in difficulty identifying where the tongue is in relation to the palate or teeth, resulting in the speech production challenges that, as noted earlier, might in turn compromise production of complex expressive language (in particular, syntax).

Two examples illustrating the value of an integrated biophysiological approach

To this point, we have described at a general level some of the implications of structural and functional characteristics of DS for swallow, speech, language, and literacy outcomes. We now briefly offer specific examples of how two of the phenotypic characteristics in DS might impact each one of the four domains of function, and offer examples of the potential clinical implications for service provision. The examples are summarized in **Figure 2** and described in the text, and illustrate the potential value of our proposed integrated approach. Some of the implications and suggestions in the figure involve reflection on language (metalinguistics, and some metacognitive skills). Such reflection may not be within the repertoire of all individuals with DS, but certainly will be within the repertoires of many of them, given that metalinguistic skills emerge at developmental age 5–7 years (Bialystok, 1986). Moreover, recent research has illustrated that when given appropriate instruction and targeted input, individuals with DS can learn and engage in metacognitive or abstract/higher order cognitive reflection (Engevik et al., 2016; O'Neill and Gutman, 2020). The framework therefore offers suggestions for targeted interventions even if individuals do not currently appear to be reflecting on their own speech.

Panel A of **Figure 2** presents examples of how difficulty in distinguishing phonemic distinctions (i.e., perceiving subtle differences in speech input) might impact each of the four functional domains. In swallowing, assessment or interventions that rely solely on auditory instructions/input might have limited value if individuals have difficulty perceiving distinctions in speech input. Consequently, visual communication supports that augment spoken input might be critically important to ensure accurate understanding of instructions for assessment or intervention in swallowing (see, for instance, Santoro, 2022). For speech, if an individual cannot discriminate between phonemes in input, they are unlikely to clearly produce those distinctions in their own spoken output, resulting in lower speech intelligibility. For language outcomes, vocabulary might be affected if individuals avoid or do not understand vocabulary words with certain phonemic distinctions; syntax might be similarly affected if phonemic distinctions that signal different morphemes are not well perceived in spoken input. Similarly, difficulty perceiving distinctions in speech input will likely interfere with literacy outcomes such as phoneme-grapheme learning, where phonemic input is matched to the graphemic representation, and/or decoding skills. Implications for service provision include making sure we assess not only hearing status, but also more specifically perceptual discrimination of speech sounds. This information can help to target services to support perceptual discrimination and to highlight instances in which limitations in vocabulary, syntax, or literacy outcomes relate not just to linguistic or cognitive challenges but may also reflect difficulty in perceiving important distinctions in spoken input.

Panel B of **Figure 2** lays out the implications for a very different phenotypic characteristic, that is, potential low oral somatosensation, on each of the four domains. In swallow, individuals who are less attuned to the coordination of their lip, tongue, and jaw will likely be less able to identify the necessary orofacial postures needed to achieve accurate or efficient functional swallowing behaviors, including controlling the bolus within the oral cavity and minimizing aspiration of food or liquid. In that event, it might be necessary to explore various food textures or liquid types (e.g., carbonated or thickened liquids), that might enhance an individual's ability to sense the food or liquid and better control swallow functioning. In speech, reduced tongue somatosensation likely influences low vowel production, due to less contact with molars compared to high vowels, resulting in lower speech intelligibility (Gick et al., 2017). Speech interventions that rely on touch cues (e.g., Hoose, 2019) will likely be less effective if individuals cannot sense the intended cues. In that event, service provision might target awareness of relative locations of jaw vs. tongue, using activities that focus on awareness of tongue and jaw location during speech, and use AAC or video modeling in that process. In language and literacy, if certain sounds are more difficult to produce due to somatosensory limitations, both vocabulary and syntax may be compromised

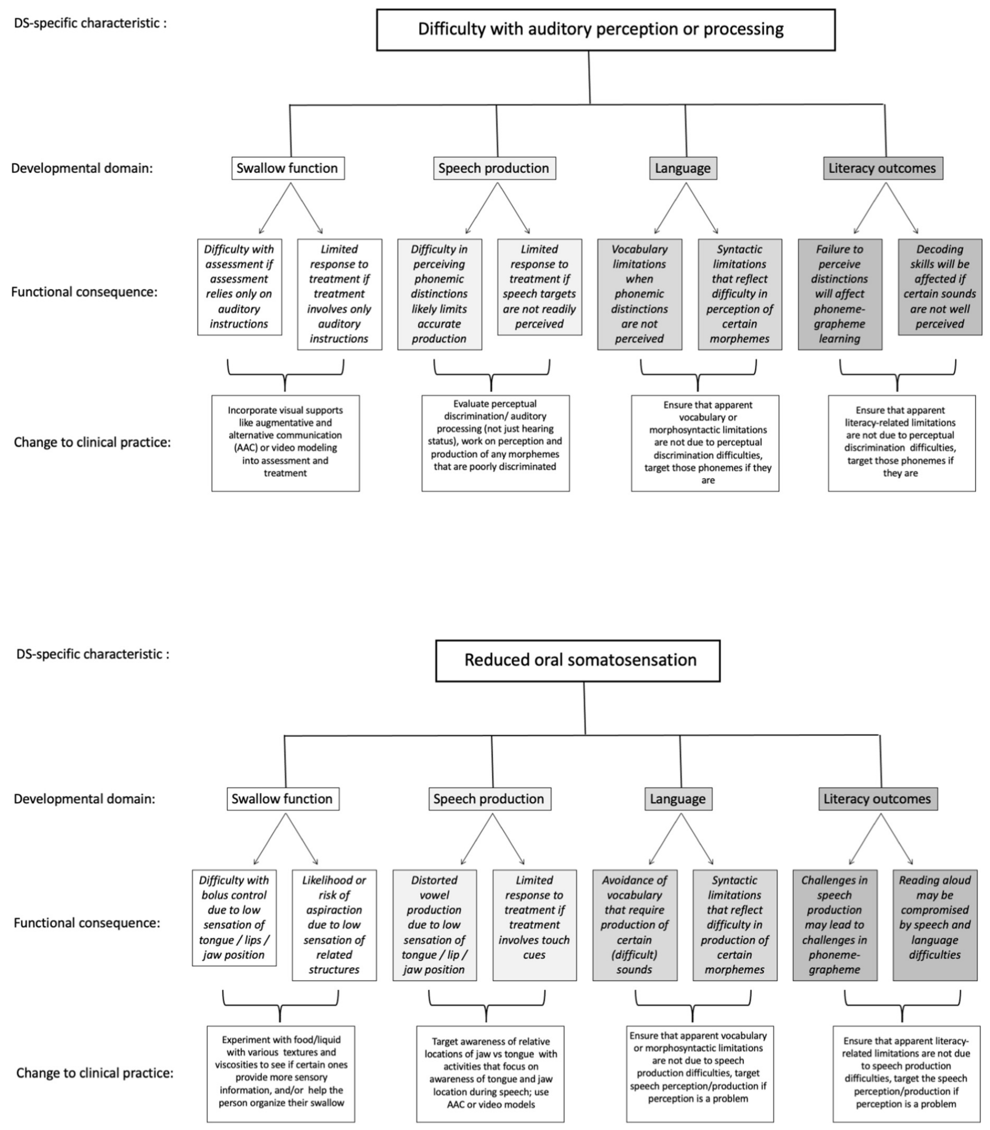

FIGURE 2
Two examples of specific ways that phenotypically linked characteristics can affect each of the four domains of swallow, speech, language, and literacy in DS, and implications for clinical practice.

due to avoidance of words or morphemes containing those sounds, and these same challenges may lead to difficulty with phoneme-grapheme acquisition as well as oral reading (reading aloud). Simply understanding that vocabulary, syntax, or literacy challenges may relate to physical difficulty with certain speech sounds will help to target interventions that include a focus on somatosensation.

Conclusion and applications to other clinical populations

A biophysiological model that combines structure, sensation, and individual factors for oropharyngeal motor activities provides an integrated approach for in-depth assessment and treatment of speech, swallowing, language, and literacy in individuals with DS. A thorough understanding of these factors and how they impact functional outcomes can be used to construct better, individualized treatment plans for individuals with DS. If one area of the model is identified as challenging to an individual, another factor within the model could be used to compensate. For example, to help achieve accurate motor movements, enhanced sensory cues for correct placement could be provided. If the individual has difficulty recognizing or processing sensory information, the therapist might increase inputs through another channel or provide feedback through multiple sensory modalities. In swallowing this can be achieved with the use of foods of various textures, stronger tastes, or even carbonation. In language and literacy, this can be achieved through multi-modal input that includes both auditory but also visual supports (see, e.g., Wilkinson and Finestack, 2021).

Although individuals with a common etiology may share similar structural and sensory changes, individual factors are important to consider, as they will vary widely. Individual factors in the assessment and treatment of swallowing could include food preferences, dietary restrictions, and nutritional needs to maintain overall health. For speech production and accuracy, individual factors could include the context in which the individual is communicating or language abilities. While not discussed in detail in this paper, the authors would like to emphasize the importance of considering these factors thoroughly while serving individuals with DS.

The authors were constrained in the amount of detail that could be provided, due to page limitations. However, many of the proposals in this article have not yet received direct research attention, and it is our hope that the outline we have provided will encourage future research on potential interrelationships. As it stands, this multidimensional, biophysiological approach to understanding complex, skilled behavior forms the basis for clinical interventions and has multiple functional implications. The purpose of this paper was to demonstrate the utilization of an adapted biophysiological framework to consider multiple dimensions that influence performance of skilled oral motor behaviors. DS was used as a clinical example to enumerate the use of the framework. However, the idea of interrelated factors in a multidimensional framework can be used with any clinical population, highlighting aspects that influence behavior in each population. For example, increased sensory processing difficulties in individuals with autism or specific neuroanatomical differences in individuals with cerebral palsy. Utilizing the framework in this way can assist in completing a holistic clinical evaluation that would aid in targeted treatment planning.

Data availability statement

The original contributions presented in this study are included in the article/supplementary material, further inquiries can be directed to the corresponding author.

Author contributions

AM and NE conceived and designed the framework presented in this study, organization, and drafting of the manuscript. LL contributed to the organization and drafting. KW provided input in conceptualizing, senior authorship, applications to DS, and drafting. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

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Home-literacy environments and language development in toddlers with Down syndrome

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Introduction: The present study aimed to (1) characterize the home-literacy environments (HLE) of toddlers with Down syndrome (DS) and (2) examine if richness of the HLE, child engagement during shared storybook reading activities, quality of a caregiver-child shared storybook reading activity, and exposure to language in the home environment predicted child receptive vocabulary concurrently (Time 1) and 6 months later (Time 2).

Methods: Participants were toddlers with DS (n = 13 at Time 1, 11–29 months of age; n = 10 at Time 2) and their mothers. Mothers completed a *Home Literacy Environment Questionnaire* at Time 1, which was used to characterize the HLE and to calculate two composite variables: richness of the HLE and child engagement in shared storybook reading. Also at Time 1, the home language environment was measured using adult word count from the LENA Recorder DLP©. The LENA was also used to audio-record and capture the quality of a caregiver-child storybook reading task in the child's home using the book *Dear Zoo*. At both time points, mothers completed the *MacArthur-Bates Communicative Development Inventories*, and the number of words understood variable was used to measure receptive vocabulary.

Results/Discussion: Results indicated that toddlers with DS experience rich HLEs and interactive shared storybook reading encounters with their mothers. A multiple linear regression revealed that child engagement and the home language environment correlated with both toddlers' concurrent and later receptive vocabularies, while the richness of the HLE and the shared storybook reading task emerged as moderate predictors of receptive vocabulary 6 months later.

KEYWORDS

home-literacy environment, shared storybook reading, early language development, Down syndrome, child word learning

1. Introduction

Down syndrome (DS) is a genetic condition caused by extra 21st chromosome material (Lejeune et al., 1959). Although there is great inter-individual variation, most individuals with DS have mild to severe intellectual disability and difficulties in speech and language (Chapman and Hesketh, 2000; Abbeduto et al., 2007). This includes delays in early language development such as first words (e.g., Abbeduto et al., 2007). Creating a rich home-literacy environment (HLE), such as providing regular access to books and participating in interactive caregiver-child shared storybook reading experiences, can have a large and positive impact on children's language and literacy skills (Dickinson and Smith, 1994; Whitehurst et al., 1994; Bus et al., 1995). However, despite the well-documented difficulties with speech and language that are common in DS (e.g., Abbeduto et al., 2007), and despite the fact that DS is associated with a unique linguistic profile (e.g., Chapman and Hesketh, 2000; Fidler, 2005; Abbeduto et al., 2007), there has been little research examining if and how HLEs might impact outcomes in this population. The purpose of the present study, therefore, was to examine the HLEs of toddlers with DS and to determine if and how the HLE impacts receptive vocabulary both concurrently and 6 months later.

The HLE is defined as children's exposure to, and the quality of, literacy-related activities in the home (DeTemple, 2001). Rich HLEs are positively related to several outcomes for young neurotypical children, including language, emergent literacy, and reading achievement (Dickinson and Smith, 1994; Whitehurst et al., 1994; Bus et al., 1995). However, richness of the HLE can be positively or negatively impacted by socioeconomic status (SES; e.g., Bus et al., 1995). For example, families with lower incomes may have less access to books and other learning materials in the home when compared to families of middle- and high-income, which in turn can negatively impact their children's language and literacy development (e.g., Neuman, 1996). The HLE is often measured by parent-reported questionnaires examining the onset, frequency, and quality of shared storybook reading, the number of books available in the home, and the frequency of trips to the library (e.g., Boudreau, 2005; Peeters et al., 2009).

Caregiver-child shared storybook reading interactions are an important component of HLEs because they expose children to more complex syntactic forms and more novel vocabulary than spoken language alone (Sulzby, 1985; Teale and Sulzby, 1986; Whitehurst et al., 1994; Cunningham and Stanovich, 1998; Pillinger and Wood, 2014). In addition, shared storybook reading provides opportunities for caregivers to scaffold their child's language development (Mason and Allen, 1986). Scaffolding is demonstrated by caregivers adapting, extending, clarifying, and/or paraphrasing stories based on their child's current level of language and literacy, thus enhancing the child's comprehension of vocabulary and story content (Altwerger et al., 1985). Other forms of scaffolding during shared storybook reading include the types of questions asked and interactive strategies that caregivers use to engage their child in the activity. For example, caregivers of young children can ask questions that require their child to point to familiar pictures in the book and/or to label something in the book, prompt their child's physical engagement (e.g., helping the child to hold the book and/or turn the pages), and/or request that their child imitate words or sound effects (Haden et al., 1996). As children develop, caregivers can transition to using more complex strategies, such as talking with their child about the story in ways that extend beyond the text, expanding on their child's responses, and even asking abstract questions that require their child to interpret, inference, or predict future story events (Haden et al., 1996). Although HLEs have often been measured via caregiver-reported questionnaires, research examining the quality of shared storybook reading for neurotypical children has more directly observed the importance of these interactions by recording and coding sessions to understand its impact on language development (e.g., Neuman, 1996).

Most studies on the HLEs of children with DS specifically have only described their HLEs via caregiver-reported questionnaires (Trenholm and Mirenda, 2006; Al Otaiba et al., 2009; van Bysterveldt et al., 2010; Ricci, 2011; Lusby and Heinz, 2020), and this body of literature has mixed findings regarding the richness of their HLEs. Some evidence suggests that young children with DS do have rich HLEs (Al Otaiba et al., 2009; van Bysterveldt et al., 2010; Lusby and Heinz, 2020; Burgoyne and Cain, 2022). For example, in two studies (Al Otaiba et al., 2009; Lusby and Heinz, 2020) with relatively large samples (n's > 100) of young children with DS (1-to-6-year-olds), a majority of caregivers reported that they began reading to their child at an early age (i.e., 1–2 years) and that they read with their child regularly (i.e., ~55–60% reported reading with their child daily for anywhere from 6 to 30 min). Further, Al Otaiba et al. (2009) reported that approximately 60% of the families in their study had 100 or more children's and adult-level books in the home. Most recently, Burgoyne and Cain (2022) noted in their study of eight caregivers of children with DS between 4 and 6 years that all caregivers reported reading to their child daily ($n = 2$) or even several times a day ($n = 6$) for 10 to 30 min. Notably, this research has reported on families from middle- to higher SES (Trenholm and Mirenda, 2006; Al Otaiba et al., 2009; van Bysterveldt et al., 2010; Lusby and Heinz, 2020; Burgoyne and Cain, 2022).

Other research has also reported rich HLEs in older, school-age children with DS from primarily middle- to high SES families. For example, using caregiver questionnaire data, van Bysterveldt et al. (2010) reported that both younger ($n = 48$ 5-to-8-year-olds) and older school-age children with DS ($n = 37$ 9-to-14-year-olds) had equal access to children's and adult-level books in the home (on average 50–75 books). Across the combined samples, 66% of the caregivers reported that they began reading to their child by 12 months of age, and 90% reported that they read to and/or with their child, though only 48% reported that this was daily (van Bysterveldt et al., 2010).

In contrast, other questionnaire-based research has found that young children, adolescents, and adults with DS from primarily middle- to upper-middle SES families are not exposed to print-rich home environments (Trenholm and Mirenda, 2006). For example, in their study of 224 caregivers of individuals with DS between 1 and 41 years, Trenholm and Mirenda (2006) reported that approximately 80% of caregivers did not read with their child daily, often spent less than 15 min reading when they did engage in shared storybook reading and had a limited number of children's books in the home. Further, although most caregivers reported reading the text and labeling pictures during shared storybook reading, only 20–30% of caregivers reported using other interactive reading strategies (e.g., asking questions about details in the story, predicting what would happen next, or asking why something happened in the story). However, this data was not broken up across the large age range, making it unclear if/how the results varied for younger versus older children, adolescents, and adults.

Only a few studies have included comparison groups, but results from these studies indicate that HLEs may not be as rich for young children with DS as they are for their neurotypical peers. For example, Ricci (2011), noted that when compared to neurotypical peers matched on chronological age, 3-to-6-year-olds with DS ($n = 20$) had less access to children's and adult-level books and less frequent caregiver-child shared storybook reading activities in the home. However, a second sample of older children with DS ($n = 17$ 8-to-13-year-olds) engaged in longer reading sessions than the younger group with DS and were explicitly taught alphabet knowledge, print knowledge, and word meanings while the younger group with DS was not (Ricci, 2011). In contrast, another questionnaire study by Westerveld and van Bysterveldt (2017) reported the HLEs of preschool-age children with DS ($n = 31$ 3-to-5-year-olds) were richer than those of children with autism spectrum disorder ($n = 80$ 3-to-5-year-olds). More than half of the caregivers (65%) from both groups reported that they began reading to their child before 1 year of age and owned at least 25 children's books, but more caregivers of children with DS (i.e., 77.4%) reported reading books "very often" with their child than did caregivers of children with autism (i.e., 33.8%). Although informative, these studies did not include children younger than 3 years old, so it is unclear if the HLEs of toddlers with DS are similar or different.

Most questionnaires regarding the HLEs of children with DS have focused specifically on caregiver behaviors, but a few studies have included questions pertaining to the child's engagement (van Bysterveldt et al., 2010; Westerveld and van Bysterveldt, 2017; Lusby and Heinz, 2020). Studies of early childhood and preschool-age children with DS suggest that they are highly engaged in shared storybook reading interactions with their caregivers. For example, Lusby and Heinz (2020) noted that roughly 70% of caregivers of younger children (1-to-6-year-olds) reported that their child "often/always" points to pictures in the book or turns the pages, and half reported that their child "often/always" names pictures in the story. However, only 33% of children "often/always" comment on the story or pictures in the book. Similarly, Westerveld and van Bysterveldt (2017) noted that caregivers of preschool-age children with DS (3-to-5-year-olds) reported that their child "often/usually" independently points to pictures in the book and/or talks about pictures in the book. In contrast, one study of elementary school-age children (van Bysterveldt et al., 2010) reported that only approximately 45% of the children in their study "often/usually" commented on pictures in the story, and only 10–30% "often/usually" asked questions about pictures, events, and/or characters in the story. However, these engagement behaviors were also more complex (e.g., asking questions) in comparison to those measured in the studies with younger children with DS (e.g., pointing to pictures). Thus, it is not clear if engagement during shared storybook reading decreases with age or if this pattern of results is simply a reflection of the types of engagement measured.

Moving beyond caregiver report, only a few studies have directly observed the quality of HLEs in DS (Fitzgerald et al., 1995; Barton-Husley et al., 2020; Burgoyne and Cain, 2022). Fitzgerald et al. (1995) examined HLEs by visiting the homes of three preschool-age children with DS twice over a 2-week period to complete a checklist of literacy artifacts (e.g., the number of children's books available in the home) and to tape-record the children's daily interactions, including caregiver-child shared storybook reading activities. These three HLEs were described as print-rich based on the number of children's books in the home (75–100) and because all mothers engaged in storybook reading activities with their child at least one time during each visit. However, only 1 of 3 mothers were observed using interactive reading strategies (e.g., labeling pictures, asking questions, expanding on the written text) during a shared storybook reading activity. Though Fitzgerald et al. (1995) extended beyond the use of survey data to measure the richness of HLEs for children with DS, the generalizability of their results is limited by the small sample size.

The remaining two studies are the only ones, to our knowledge, that both directly observed HLEs of children with DS and examined if and how HLEs and shared storybook reading impacted language and literacy development in their samples. Burgoyne and Cain (2022) directly observed shared storybook reading interactions between eight children with DS (aged 4–6 years) and their parents by visiting the children's homes to video record two separate mother–child storybook reading activities in the home using two different books. One book was to be read in its original form to illustrate a typical shared storybook reading interaction; the second book was modified to include embedded prompts (i.e., picture labeling, vocabulary, linking text to general knowledge, and inferencing). Parents used more extra-textual talk (i.e., asking questions, commenting, responding to their child) when they read the book with embedded prompts than during the typical shared storybook reading activity with their child. Further, children with DS showed greater participation in reading and produced significantly more words ($M = 110.63$, $SD = 78.07$) and a greater range of words ($M = 47.13$, $SD = 25.0$) when reading books with embedded prompts compared to when reading books during a typical reading session (words produced $M = 52.50$, $SD = 35.84$; different words $M = 27.13$, $SD = 17.28$; Burgoyne and Cain, 2022). Although the sample was small, these data support that more interactive reading strategies can promote language development in young children with DS.

Lastly, Barton-Husley et al. (2020) directly observed the use of interactive reading strategies between 22 mothers with children with DS (22-to-63-months old) compared to 22 mothers of neurotypical children matched on chronological age. The quality of the HLEs was examined by visiting the children's homes once for 1 to 2 h to video record a mother–child shared storybook reading activity in the home. During shared storybook reading activities, mothers of children with DS were observed using interactive reading strategies (i.e., questions, descriptions, gestures, and labels) more frequently than did the mothers of the neurotypical peers (73% vs. 50% of recorded sessions). Further, their results indicated that mothers of children with DS who used a fewer total number of utterances during a shared storybook reading activity had children with higher receptive language raw scores as measured by the *Mullen Scales of Early Learning* (Mullen, 1995), indicating that mothers of children with DS adapt their language to meet the language and engagement needs of their child. For example, it is possible that mothers of children with DS with higher receptive language skills talked less because they were giving their child more time to talk, whereas mothers of children with DS with lower receptive language skills talked more to help scaffold language development. Though Barton-Husley et al. (2020) were the first to examine the relationship between maternal input and receptive vocabulary in toddlers and preschool-age children with DS, the results of their study are limited to a single time point. More research, particularly longitudinal research, is needed to understand the impact of HLEs and shared storybook reading on the word learning of young children with DS.

Although the benefits of rich HLEs and interactive shared storybook reading have been well documented in the neurotypical literature (e.g., Bus et al., 1995; Neuman, 1996), there is limited research examining if and how these impact language and literacy development for children with DS. There is a clear need to examine the HLEs of toddlers with DS and to determine if the richness of the HLEs impacts early language development concurrently and/or over time. Further, to fully capture the quality of shared storybook reading, it is necessary to go beyond caregiver report and include observational data to measure these interactions more directly. The purpose of the current study was to describe the HLEs of toddlers with DS, observe and document the quality of a shared storybook reading task between toddlers with DS and their mothers, and measure the relation between these variables and word learning in toddlers with DS. Specifically, the research questions for this current study were:

a. How do mothers of toddlers with DS describe their HLEs using a parent questionnaire? Further, how do mothers of toddlers with DS report their child's engagement during shared storybook reading activities using a parent questionnaire?
b. What interactive strategies do mothers of toddlers with DS use during a shared storybook reading task (i.e., what is the quality of a shared storybook reading task)?
c. Do differences in the richness of HLEs and quality of parent–child shared storybook reading relate to receptive language outcomes concurrently and 6 months later?

2. Materials and methods

2.1. Participants

Participants included toddlers with DS and their mothers who were part of a larger study examining early language development in DS. For the present study, participant data was used from two time points, referred to as Time 1 and Time 2, once the shared storybook reading task had been added to the protocol. Thirteen families participated in the study at Time 1. One of those participants did not complete the shared storybook reading task with their child because the battery in the LENA Recorder DLP© sent to them died due to delayed participant compliance in completing the task. Data from 10 families were included for follow-up testing approximately 6 months later ($M = 7.30$ months, $SD = 4.42$ months) at Time 2. One participant did not have data at Time 2 as they had already completed and exited the overall study at this time, and two families were non-responsive at Time 2. One participant completed their follow-up testing 19 months later at Time 2 due to COVID-19, which paused the study in March 2020. Participants were 11–29 months at Time 1 ($M = 17.92$, $SD = 5.27$), and 16–48 months at Time 2 ($M = 24.30$, $SD = 9.08$). Additionally, 76.9% of the participants were white, and 69.2% were males. Mothers reported an annual family income ranging from $20,000 to > $300,000. Of the mothers in this study, 30.8% had attended some college, 23.1% were college graduates, 7.7% had postgraduate training, and 38.5% had a professional degree (e.g., MA, PhD). Participants were recruited from the Midwest and Southeast regions of the United States using emails and posting on social media to local DS organizations and early intervention service organizations. As reported by the participants' mothers, all children had normal or corrected hearing and vision, and English was the primary language spoken in the home. Additional participant descriptive statistics are reported in Table 1.

TABLE 1 Participant descriptive statistics.

Variable	*M*	*S.D.*	Range
Chronological age, child[a]	17.92	5.27	11–29
Chronological age, mother[b]	33.85	5.86	21–46
Early learning composite[c]	62.46	10.25	49–82
Words understood[d]			
Time 1	100.15	82.67	0–301
Time 2	138.10	114.97	6–309
Words produced[d]			
Time 1	6.08	7.54	0–25
Time 2	15.10	21.02	0–66
	f	%	
Family income			
<$50,000	2	15	
$50,001–$100,000	6	46	
$100,001–$150,000	3	23	
$150,001–$200,000	0	0	
$200,001–$250,000	0	0	
$250,001–$300,000	0	0	
>$300,000	1	8	
Not reported	1	8	

[a] Child's age is reported in months at Time 1.
[b] Mother's age is reported in years at Time 1.
[c] Composite scores derived from *Mullen Scales of Early Learning* (MSEL) at Time 1.
[d] Raw scores derived from *MacArthur-Bates Communicative Development Inventories* (CDI).

2.2. Procedures

In-person visits were conducted at both time points in the family's home or at a location nearby (e.g., library, DS community center). As part of a larger-assessment battery, the *MacArthur-Bates Communicative Development Inventories* (CDI) was completed by the child's mother, and the *Mullen Scales of Early Learning* (MSEL) was administered to the child by a trained examiner. Families were also provided with a *Home Literacy Environment Questionnaire* and a LENA Recorder DLP© with instructions for a shared storybook reading task using the book *Dear Zoo* (Campbell, 1982).

2.3. Measures

2.3.1. Home-literacy environment questionnaire

For this study, we created an HLE questionnaire (adapted from Boudreau, 2005; Peeters et al., 2009; van Bysterveldt et al., 2010; 5 min; see Supplementary material) to characterize the richness of the HLE and child engagement in reading. The questionnaire included short answer ($n = 3$), Likert-type ($n = 6$), and forced choice ($n = 19$) questions. From this questionnaire, two composite variables were calculated and used in the regression analyses (research question 3): (1) parent-reported richness of the HLE and (2) parent-reported child engagement in shared storybook reading.

2.3.1.1. Richness of the HLE

The composite for parent-reported richness of the HLE was comprised of 13 questions pertaining to the exposure and nature of literacy-related activities in the home, including accessibility of children's and adult-level books in the home, parental perspectives on the importance of reading with children, amount of time spent reading, number of books read to their children, and interactive reading styles utilized during a typical shared storybook reading interaction. The sum of the 13 items was used as an indicator of the richness of the HLE, with higher scores representing richer HLEs (highest possible score: 67).

2.3.1.2. Child engagement

The composite for child engagement in shared storybook reading was comprised of eight questions related to what the child does during a typical shared storybook reading interaction. The sum of the eight items was used as an indicator of child engagement in reading, with a higher score representing greater interest and engagement in shared storybook reading (highest possible score: 36).

2.3.2. Language environment analysis recorder and software©

The LENA Recorder DLP© and LENA software are a system for analyzing the language environment of a child in their day-to-day life (LENA Research Foundation, 2018). The LENA Recorder DLP© is a small, wearable recorder that records for up to 16 h (when children are napping or bathing, caregivers are instructed to leave the recorder on nearby). From these recordings, adult word count, child vocalizations, and conversational turns can be extracted using the LENA PRO© or SP© software (LENA Research Foundation, 2018). The normative sample of the LENA software is based on audio of 2-to-48-month olds from families of varying socioeconomic backgrounds (Gilkerson et al., 2008), and norms were developed from recordings captured in a 12 h long, spontaneous speech environment (Gilkerson et al., 2008; LENA Research Foundation, 2018).

2.3.2.1. Home language environment

In the present study, the home language environment was measured using the adult word count (AWC) from the LENA Recorder DLP©, which is automatically calculated by the LENA software.

2.3.2.2. Shared storybook reading task

For the shared storybook reading task, caregivers were instructed to read the age-appropriate, lift-the-flap book, *Dear Zoo* (Campbell, 1982) during any normal reading time in a quiet environment when the child was wearing the LENA Recorder DLP©. The book contains predictive text and several unfinished sentences noted by an ellipsis to prompt the reader(s) to open the flap and label the pictured animal (i.e., *So they sent me a*…). Mothers were asked to note the date and time that they read the story, so that this interaction could be identified in the LENA recording for transcription. *Systematic Analysis of Language Transcription* (SALT; Miller and Iglesias, 2006) software and conventions were used to transcribe the samples and then coded using an adapted coding scheme to measure the quality of the caregiver-child shared storybook reading task, including the use of maternal interactive reading styles (i.e., Neuman, 1996; Crowe, 2000; Justice et al., 2003; McDuffie et al., 2018). Specifically, all maternal reading behaviors were coded for labels, expansions, questions, comments, requests, reading written text verbatim, paraphrasing the story, repetition of questions, and non-story speech. A composite variable was calculated to represent the quality of caregiver-child shared storybook reading for use in regression analyses (research question 3, see details below).

2.3.2.2.1. Transcriber training

The primary transcriber, the first author, was trained using SALT software conventions (Miller and Iglesias, 2006). Training also included independent transcription of practice language samples from a different mother–child shared storybook task with children with DS. Each practice language sample transcript was compared to a standard transcript at the utterance level for all maternal utterances across multiple transcription dimensions. Once the primary transcriber transcribed two consecutive transcripts with at least 70% agreement for utterance segmentation, word identification, and number of morphemes and words and at least 80% agreement on the dimensions of unintelligibility, abandoned utterances, mazes, overlapping speech, and ending punctuation, they were considered trained to fidelity.

2.3.2.2.2. Transcription of shared storybook reading task

Each shared storybook reading task was independently transcribed by the primary transcriber and then independently checked by another transcriber trained on SALT software conventions. Maternal utterances were segmented into Communication units (C-Units; i.e., an independent clause and any modifiers, which could include a dependent clause), recommended for individuals over 3 years (Loban, 1976). There were 87 discrepancies between the primary transcriber and checker. These were reconciled by the primary transcriber.

2.3.2.2.3. Coding of shared storybook reading task and outcome variables

The quality of the shared storybook reading task was measured via an adapted coding scheme (i.e., Neuman, 1996; Crowe, 2000; Justice et al., 2003; McDuffie et al., 2018). This adapted coding scheme focused on the use of maternal reading behaviors (i.e., labels, expansions, questions, comments, requests, reading written text verbatim, paraphrasing the story, repetition of questions, and non-story speech). All maternal reading behaviors during the shared storybook reading task were coded and summed to create a total maternal utterances variable. Maternal utterances that were coded as labels, expansions, questions, comments, and requests were considered "interactive" reading strategies. Reading the written text verbatim, paraphrasing the text, repetition of questions, and non-story speech were considered codable utterances but were not counted as interactive reading strategies. Coded maternal reading behaviors and examples of each can be found in Table 2.

Three primary outcome variables were calculated from the shared storybook reading task. First, the number of mothers who used each interactive strategy were counted. Second, the percentage of each of the interactive reading strategies used by the mothers was calculated (i.e., percentage of total maternal utterances that were labels, expansions, questions, comments, or requests). Third, a shared storybook reading (SBR) composite score was calculated to represent the quality of the task for use in regression analyses. An SBR was calculated for each participant by dividing their total number of utterances that were interactive

TABLE 2 Maternal reading behaviors during the caregiver-child shared storybook reading task.

Code	Definition	Example
*Labels	Label of a pictured agent or object following a spontaneous question (i.e., a question not included in the text) that typically elicited a one-word naming response	Mother: "What is it?" "Giraffe!"
*Expansions		
Comment	Added information that was outside the written text and/or comments on the pictures	Mother: "The giraffe is in the box."
Bridge	Making connections from story content to everyday experiences	Mother: "We went to the zoo and saw a tall giraffe."
*Questions		
Yes/No	Question that required a "yes" or "no" response from the child	Mother: "Do you see the giraffe?"
Tag	Question that could be answered by "yes" or "no" but typically was rhetorical or served as a form of commenting	Mother: "He's in a box, is not he?"
Label	Question that typically elicited a one-word naming response	Mother: "What is that?"
Descriptive	Question that required a response to describe an action related to the story	Mother: "What is happening in this picture?"
Complex	Question that required a semantically complex response (i.e., predictive, casual)	Mother: "What will happen next?"
Choice	Question that required a choice from the child	Mother: "Is the giraffe big or little?"
*Comments	General comment that related to the shared storybook reading activity	Mother: "I really like this story."
*Requests		
Imitative	Request for imitation	Mother: "Say, 'giraffe'!"
Sound effects	Request for sound effect	Mother: "What does a lion say?"
Reading text verbatim	Utterance that was verbatim from the written text	Mother (reading text): "I wrote to the zoo to send me a pet."
Text completion	Label (i.e., one-word naming response) of a pictured agent or object that completed the written text, "*So they sent me a(n)…*"	Mother (reading text): "So they sent me a…" Mother (opens flap to see picture): "Giraffe!"
Paraphrasing	Utterance that paraphrased the story but was not reading the written text verbatim	Text: "They sent me an…" Paraphrase: "And here's what they sent me."
Repetition of questions	Utterance that repeated child's utterance as a question	Child: "Giraffe." Mother: "Giraffe?"
Non-story speech	An utterance that did not contain story content or did not relate to the reading task (e.g., side conversation or behavior management: attention, non-desirable, praise)	Mother: "Stop doing that."

*Denotes that these codes were considered "interactive" reading strategies when calculating the SBR composite score measuring the quality of the caregiver-child shared storybook reading task for use in regression analyses; research question 3.

strategies by their total number of maternal utterances. Then, the SBR composite was calculated by averaging individual SBR scores across participants.

2.3.2.2.4. Coder training

The primary transcriber also served as the primary coder. First, the primary coder discussed the adapted coding scheme with a second, reliability coder by providing definitions and examples of each code. The primary coder then shared a coded transcript with the reliability coder and discussed each coding choice.

2.3.2.2.5. Coding reliability

After coder training, each shared storybook reading task was independently coded by the primary coder and independently reviewed by the reliability coder. There were 21 discrepancies in coding. These were reconciled by the primary coder.

2.3.3. Vocabulary

The *MacArthur-Bates Communicative Development Inventories-Words & Gestures* (CDI; Fenson et al., 2007) was used to measure the children's receptive vocabulary (i.e., words understood) at both Time 1 and Time 2. The CDI is a standardized parent-reported checklist designed to assess children's receptive and expressive vocabulary skills from 8 to 30 months. The CDI has good reliability and validity (Fenson et al., 2007). The internal consistency coefficients for the vocabulary scales range from 0.95 to 0.96, and test–retest reliability coefficients for Words and Gestures range from 0.61 to the mid 0.80s. The CDI Words and Gestures correlates (i.e., has concurrent validity) with the *Expressive One-Word Picture Vocabulary Test* (Gardner, 1981), with correlations ranging from 0.73 to 0.86 and with the *Reynell Developmental Language Scales*, *Expressive* (Reynell and Gruber, 1990), with correlations ranging from 0.75 to 0.82. The CDI has previously been used with children with DS, and moderate to strong correlations have been reported between the CDI and other measures of vocabulary (0.70; Miller et al., 1995) and language (0.77; i.e., *Bayley Scales of Infant Development*; Heilmann et al., 2005) for children with DS. Words understood was used as the outcome variable. Words produced was not used in this study because very few participants were using spoken language at this time.

2.3.4. Cognitive abilities

The *Mullen Scales of Early Learning* (MSEL; Mullen, 1995) was administered by a trained examiner to measure the children's overall cognitive abilities at Time 1. The MSEL is a standardized assessment tool designed to measure development from birth to 68 months of age across four domains, Visual Reception, Fine Motor, Receptive Language, and Expressive Language, which together yield an overall measure of cognitive abilities, the Early Learning Composite (ELC). Like other standardized, norm-referenced assessments of cognition (used to estimate level of intellectual abilities), the ELC has a mean of 100 and a standard deviation of 15. In addition, an overall developmental age can be calculated by averaging the age equivalents from these four domains. The internal consistency coefficients range from 0.83 to 0.95, test–retest reliability coefficients range from 0.82 to 0.85, and interrater reliability coefficients range from 0.91 to 0.99 (Mullen, 1995). The MSEL has strong concurrent validity with other standardized tests of early child development and cognition [e.g., *Bayley Scales of Infant Development* (Bayley, 1993), *Peabody Developmental Motor Scales* (Folio and Fewell, 1983), *Birth to Three Developmental Scale* (Dodson and Bangs, 1979)]. Additionally, content validity and construct validity have been established (Mullen, 1995).

2.4. Data analysis plan

For research question 1, descriptive statistics were used to report how mothers of toddlers with DS characterize their HLEs, use of interactive reading styles, and child engagement via the HLE questionnaire. For research question 2, descriptive statistics were again used to describe the quality of a shared storybook reading activity between mothers and their child with DS via coding of a caregiver-child shared storybook reading task. For research question 3, multiple linear regression was used to examine if the richness of parent-reported HLEs, parent-reported child engagement in reading activities, the quality of a caregiver-child shared storybook reading task, and the home language environment predicted receptive language outcomes at Time 1 and Time 2. Words understood at Time 1 was slightly skewed (Skewness = 1.12), and several variables had slightly elevated kurtosis values (words understood Time 1 = 1.55, words understood Time 2 = −1.50, child engagement = −1.17, adult word count = −1.20). For the regression predicting words understood at Time 1, there were no serious violations of the assumptions of multiple regression, including multicollinearity. For the regression predicting words understood at Time 2, there were two participants identified as having standardized residuals above 3.0 or below −3.0. These were the participants with the highest and lowest words understood scores, 6 (this was the youngest participant) and 158, respectively. Because the sample size was already reduced at Time 2, and because we believed these scores reflected each participant's true receptive vocabulary abilities, we elected to keep these participants in the analyses. There were no other major violations of multiple regression, including multicollinearity. Follow-up analyses were also run adding age to the regression models as a control variable. Given the small sample size and that we already had four predictors (i.e., our primary predictors of interest: HLE, caregiver-child shared storybook reading, child engagement in shared storybook reading, and the home language environment), we added age as a predictor after running the originally proposed regressions. Further, because participants had younger developmental ages, we consider the regressions with age exploratory and suggest caution in interpreting these findings. Further, given that one participant was not able to complete the Time 2 session until 19 months later (see participant information above), the Time 2 regression analyses were run both with and without this participant included. The pattern of results did not change, and the statistical results themselves changed only minimally. Thus we retained this participant in our presented results. Descriptive statistics of key variables can be found in Table 3. Correlations among key variables can be found in Table 4.

3. Results

3.1. Research question 1: home-literacy environment, Time 1

On the HLE questionnaire, all mothers reported that they began reading to their child between pregnancy and 12 months of age ($M = 1.88$, $SD = 4.05$), and approximately half ($n = 6$) had a designated reading time with their child. Mothers also reported that they had many children's ($M = 122.31$, $SD = 99.26$, *range* = 30–300) and adult-level books ($M = 120.77$, $SD = 74.77$, *range* = 20–200) in their home. Most mothers ($n = 11$) reported reading with their child regularly, and when asked about different interactive reading strategies during caregiver-child shared storybook reading, a majority agreed (i.e., "strongly agree"/ "agree") that they use interactive strategies during typical storybook reading activities with their child ($n = 13$ point out details outside the written text; $n = 10$ relate the story to the child's experiences; $n = 10$ teach their child letters and sounds). The exception was that only half ($n = 7$) agreed that they ask their child questions about the story and follow-up with answers. Frequency of mother responses to additional items on the HLE questionnaire can be found in Table 5.

When asked about their child's engagement in shared storybook reading activities, approximately half ($n = 6$) reported that their child asks to read books or pretends to read the story in a book (e.g., sitting with a book and producing speech that is similar to the actual story in the book). When asked questions pertaining to the child's engagement during book reading, most mothers reported that their child interacts with the story by grabbing for and/or holding the book ($n = 13$), turning the pages ($n = 12$), and pointing to pictures or words on the page ($n = 8$). In contrast, most mothers reported that their child does not regularly name familiar pictures in the book ($n = 12$), ask questions about the story ($n = 13$), or fill in words or lines of a familiar story ($n = 13$). Mothers' responses to items on the HLE

TABLE 3 Descriptive statistics of key variables.

Variable	*M*	*S.D.*	Range	*N*
HLE[a]	34.89	8.12	21.0–49.0	13
Child engagement[b]	12.38	6.38	4.0–23.0	13
Home language environment (AWC)[c]	17,142.62	8,867.64	4,865–30,675	13
SBR[d]	0.35	0.12	0.20–0.60	12

[a]Composite score derived from the HLE questionnaire to describe the richness of the HLE.
[b]Composite score derived from HLE questionnaire to describe child engagement during typical shared storybook reading activities.
[c]Adult word count (AWC) derived from LENA Recorder DLP© to measure home language environment.
[d]Composite score representing the average quality of the shared storybook reading task across participants.

TABLE 4 Correlations among key variables.

Variable	1	2	3	4	5	6	7	8
1. Child age	–							
2. HLE	0.42	–						
3. Child engagement	0.77**	0.32	–					
4. Home language environment (AWC)	−0.19	−0.21	0.07	–				
5. SBR	0.07	−0.08	0.11	0.18	–			
6. WU Time 1	0.83***	0.41	0.82***	−0.40	−0.08	–		
7. WU Time 2	0.79**	0.52	0.89***	−0.23	0.29	0.89***	–	
8. ELC	−0.37	−0.54^{+}	0.05	0.25	0.16	−0.27	−0.08	–

HLE, Richness of HLE; AWC, Adult Word Count; SBR, Quality of Shared-Book Reading Task; WU Time 1, Words Understood at Time 1; WU Time 2, Words Understood at Time 2; ELC, Early Learning Composite derived from *Mullen Scales of Early Learning* (MSEL).
$^{+}p < 0.07$, $^{**}p < 0.01$, $^{***}p < 0.001$.

questionnaire related to child engagement in reading can be found in Table 6.

3.2. Research question 2: shared storybook reading task, Time 1

Frequency counts of the number of mothers who used each interactive shared storybook reading strategy are reported below. Means, standard deviations, ranges, and percentages of all maternal coded reading behaviors at Time 1 are presented in Table 7. Expansions, questions, and labels made up the majority of the interactive reading strategies used. Requests and comments were used less often.

3.2.1. Labels

Ten of the 12 mothers used labels by asking spontaneous questions (e.g., "What is that?) that they then followed with a one-word naming response (e.g., "Giraffe!").

3.2.2. Expansions

All 12 mothers expanded on the written text and/or commented on the pictures during the shared storybook reading task. Seven of the 12 mothers used expansion-bridges by connecting the story content to the child's everyday experiences during the shared storybook reading task.

3.2.3. Questions

Seven of the 12 mothers asked yes-or-no questions during the shared storybook reading task. Ten of the 12 mothers asked labeling questions during the shared storybook reading task. Two mothers asked descriptive questions, and two mothers asked complex questions during the shared storybook reading task. Only one mother asked a tag and/or a choice question during the shared storybook reading task.

3.2.4. Comments

Eight of the 12 mothers made general comments related to the shared storybook reading task.

3.2.5. Requests

Three of the 12 mothers requested that their child imitate a word during the shared storybook reading task. Similarly, three mothers requested that their child make sound effects during the shared storybook reading task.

3.3. Research question 3: predicting child word learning

3.3.1. Time 1

Multiple linear regression was conducted with parent-reported richness of the HLE (via the HLE composite from the HLE questionnaire), parent-reported child engagement in book reading (via the child engagement composite from the HLE questionnaire), quality of the shared storybook reading task (via the SBR composite from the LENA), and the home language environment (via AWC from the LENA) as predictors of child receptive vocabulary at Time 1. A significant model emerged, $F(4, 7) = 15.10$, $p = 0.001$, with approximately 90% of the variance in child receptive vocabulary accounted for by the linear combination of predictors (see Table 8). Examination of the predictor variables showed significant, unique effects, with child engagement explaining 63% unique variance and home language environment (AWC) explaining 17% unique variance. See the Supplementary materials for scatterplots of the data.

3.3.1.1. Age

A follow-up analysis was conducted to examine the role of age in our regression model. Thus, we performed the analysis again but added age as a control variable. When age from Time 1 was entered alone, it significantly predicted child receptive language at Time 1, $F(1, 10) = 21.84$, $p < 0.001$, and the R^2 indicated that 69% of the variance in child receptive language was accounted for by age (see Table 9). When the other variables were added to the model, the model remained significant, $F(5, 6) = 13.31$, $p = 0.003$, with approximately 92% of the variance in child receptive vocabulary accounted for by the linear combination of predictors. This accounted for an additional 23% of the variance in child receptive language, *F change* $(4, 6) = 4.20$, $p = 0.06$. Examination of the predictor variables in Step 2 indicated that age was no longer a significant predictor, but child engagement explained 16% unique variance and home language environment (AWC) explained 12% unique variance.

TABLE 5 Frequency of mothers' responses to items on HLE questionnaire related to the richness of HLE.

Question	Never/0	1–2	3–4	5–6	7–8	9–10	11+
How many times in a week mother reads books for enjoyment	3	4	4	0	1	1	0
How many times in a week mother reads informative books	3	7	1	2	0	0	0
How many times in a week mother reads to child	0	0	2	3	2	3	3
How many books mother read to child in past week	0	1	0	4	2	1	5
How many books mother reads to child in one sitting	0	5	7	1	0	0	0
	<15 min	**15–30 min**	**30–45 min**	**1–2 h**	**3–4 h**	**5–6 h**	**7+ h**
How much time mother spent reading to child in past week[a]	0	0	4	3	4	1	0
	<10 min	**10–20 min**	**21–30 min**	**31–40 min**	**41–50 min**	**51–60 min**	**>1 h**
How much time mother spends reading to child in one sitting	2	8	2	1	0	0	0
	Never	**Once**	**Every 4–6 mos**	**Every 2–3 mos**	**Monthly**	**Every 2–3 wks**	**Weekly**
How often mother took child to library/bookstore in the last year	1	3	2	0	0	4	3

Interactive book reading styles	Strongly disagree	Disagree	Agree	Strongly agree
Points out details outside of written text	0	0	6	7
Relates the story to child's everyday experiences	0	3	4	6
Asks questions and follows-up with answers	2	4	7	0
Teaches child letters/sounds of letters	1	2	7	3

N = 13. Min, Minutes; H(s), Hour(s); Wks, Weeks; Mos, Months.

[a] One participant did not report an answer.

TABLE 6 Frequency of mothers' responses to items on HLE questionnaire related to child engagement.

Question	Never	1–2	3–4	5–6	7–8	9–10	11+
How many times child asks mother to read to him/her in a week	7	2	1	1	0	0	2
How many times child pretends to read books in a week[a]	6	1	0	1	2	0	2

"During storybook reading time with your child, does he/she…"	Never	Has but rarely	Occasionally	A few times per story	Frequently during story
Grabs for/holds book	0	0	3	1	9
Turns pages of book	1	0	2	1	9
Points to pictures/words in book independently	5	0	3	2	3
Names familiar pictures	9	3	0	1	0
Asks questions about story	13	0	0	0	0
Fills in words/lines of a familiar story	13	0	0	0	0

N = 13.

[a] One participant did not report an answer.

TABLE 7 Use of interactive reading strategies by type during the caregiver-child shared storybook reading task.

Variable	Total number	*M*	*S.D.*	Range
Maternal utterances	801	66.75	23.14	43–116
Utterances that were interactive strategies	297	24.75	16.97	9–70
	Total number (% of maternal utterances)	***M***	***S.D.***	**Range**
*Labels	69 (8.61)	5.75	5.71	0–21
*Expansions				
Comment	87 (10.86)	7.25	5.26	2–19
Bridge	13 (1.62)	1.08	1.08	0–3
*Questions				
Yes/No	17 (2.12)	1.42	1.68	0–5
Tag	1 (0.12)	0.08	0.29	0–1
Label	65 (8.11)	5.42	5.55	0–19
Descriptive	4 (0.50)	0.33	0.89	0–3
Complex	3 (0.37)	0.25	0.62	0–2
Choice	1 (0.12)	0.08	0.29	0–1
*Comments	17 (2.12)	1.42	1.56	0–5
*Requests				
Imitative	14 (1.75)	1.17	2.59	0–8
Sound effects	6 (0.75)	0.5	1.17	0–4
Reading text verbatim	323 (40.32)	26.92	4.19	21–37
Text completion	47 (5.87)	3.92	2.78	0–8
Paraphrasing	15 (1.87)	1.25	1.91	0–6
Repetition of questions	9 (1.12)	0.75	1.06	0–3
Non-story speech	110 (13.73)	9.17	6.94	1–28

N = 12. *Were considered "interactive" reading strategies when calculating the composite score measuring the quality of the caregiver-child shared storybook reading task for use in regression analyses; research question 3.

TABLE 8 Multiple linear regression predicting receptive vocabulary at Time 1.

Predictor variable	Beta	*t*	*p*
HLE	0.04	0.27	0.79
Child engagement	0.85	6.51	<0.001
SBR	−0.10	−0.76	0.47
Home language environment (AWC)	−0.44	−3.42	0.01

N = 12. HLE, Richness of HLE; SBR, Quality of Shared-Book Reading Task; AWC, Adult Word Count.

3.3.2. Time 2

Multiple linear regression was conducted to examine if Time 1 measures (parent-reported richness of the HLE, parent-reported child engagement in book reading, quality of the shared storybook reading task, and the home language environment) predicted child receptive vocabulary at Time 2. A significant model emerged, $F(4, 5) = 119.80$, $p = < 0.001$, with approximately, 99% of the variance in child receptive vocabulary at Time 2 accounted for by the linear combination of predictors (see Table 10). Examination of the predictor variables showed a significant, unique effect for all predictors: richness of the HLE (4% unique variance explained), child engagement (57%), quality of the shared storybook reading task (SBR; 7%), and home language environment (AWC; 8%). See the Supplementary materials for scatterplots of the data.

3.3.2.1. Age

Again, we conducted a follow-up analysis to examine the role of age, with age added to our regression model as a control variable. When age at Time 1 was entered alone, it significantly predicted child receptive language at Time 2, $F(1, 8) = 13.16$, $p = 0.007$, and the R^2 indicated that 62% of the variance in child receptive language at Time 2 was accounted for by age at Time 1 (see Table 11). When the other Time 1 measures were added to the model, the model remained significant, $F(5, 4) = 12.06$, $p = 0.016$, with approximately 94% of the variance in child receptive vocabulary accounted for by the linear combination of predictors. This accounted for an additional 32% of the variance in child receptive language, *F change* $(4, 4) = 5.08$, $p = 0.07$. Examination of the predictor

TABLE 9 Multiple linear regression predicting receptive vocabulary at Time 1 accounting for age at Time 1.

Predictor variable	*B*	Beta	*t*	*p*
Model 1				
Age	13.14	0.83	4.67	<0.001
Model 2				
Age	4.06	0.26	1.27	0.25
HLE	0.06	0.01	0.05	0.97
Child engagement	8.74	0.65	3.40	0.01
SBR	−74.47	−0.11	−0.87	0.42
Home language environment (AWC)	−0.003	−0.38	−2.90	0.03

$R^2 = 0.66$ for Model 1; R^2 change = 0.23 for Model 2; Total $R^2 = 0.92$. HLE, Richness of HLE; SBR, Quality of Shared-Book Reading Task; AWC, Adult Word Count.

TABLE 10 Multiple linear regression predicting receptive vocabulary at Time 2.

Predictor variable	Beta	*t*	*p*
HLE	0.23	4.53	0.006
Child engagement	0.81	16.67	<0.001
SBR	0.27	5.86	0.002
Home language environment (AWC)	−0.29	−6.10	0.002

N = 10. HLE, Richness of HLE; SBR, Quality of Shared-Book Reading Task; AWC, Adult Word Count.

TABLE 11 Multiple linear regression predicting receptive vocabulary at Time 2 accounting for age at Time 1.

Predictor variable	*B*	Beta	*t*	*p*
Model 1				
Age Time 1	16.20	0.79	3.63	0.007
Model 2				
Age Time 1	−2.69	−0.13	−0.51	0.64
HLE	3.09	0.24	1.77	0.15
Child engagement	15.20	0.93	3.72	0.02
SBR	121.62	0.10	0.79	0.48
Home language environment (AWC)	−0.003	−0.26	−1.73	0.16

$R^2 = 0.62$ for Model 1; R^2 change = 0.32 for Model 2; Total $R^2 = 0.94$. HLE, Richness of HLE; SBR, Quality of Shared-Book Reading Task; AWC, Adult Word Count.

variables in Step 2 indicated that age no longer contributed unique significant effects. Further, richness of the HLE, quality of a shared storybook reading task, and the home language environment (AWC) did contribute to the overall model when age was *not* in the model (see Table 10), this was no longer the case once age was entered into the model. Nonetheless, child engagement remained significant, explaining 22% of the variance when age was in the model.

4. Discussion

The purpose of the current study was to examine the HLEs of toddlers with DS and determine if and how the HLE impacts their early word learning. Specifically, we collected data to describe the HLEs of toddlers with DS, documented the quality of a shared storybook reading task between those toddlers and their mothers, and measured the relationship between these variables and early language development. Although HLEs have been documented as important predictors of language and literacy outcomes in neurotypical children (e.g., Dickinson and Smith, 1994; Whitehurst et al., 1994; Bus et al., 1995), there has been limited research on the HLEs of children with DS. Further, our study added to the existing DS literature by examining child engagement during caregiver-child shared storybook reading via the use of an HLE questionnaire, directly measuring the quality of a shared storybook reading task via the use of a LENA recorded language sample, and including a longitudinal component to examine early language in toddlers with DS. Overall, our results indicate that some toddlers with DS, at least those from relatively higher socioeconomic status families, have rich HLEs and that child engagement and the home language environment correlate with their concurrent and later receptive vocabulary abilities. Richness of the HLE and quality of the shared storybook reading task also emerged as moderate predictors of word learning 6 months later. Interestingly, while age alone correlated with both concurrent and later child receptive vocabulary, once variables related to HLE and shared storybook reading were added to regression models, age was no longer a significant correlate. Below we review our findings in more detail and discuss their implications for caregivers and practitioners.

4.1. Richness of the HLE

Our first research question was how mothers of toddlers with DS characterize their HLEs and book reading styles. The caregivers in our study indicated that on average there were 100 children's books in the home. Further, more than half of the mothers reported that they read with their child 7–11 times a week and spent 10–30 min reading together per session. These findings are consistent with previous research suggesting rich HLEs for young children with DS (Al Otaiba et al., 2009; van Bysterveldt et al., 2010; Lusby and Heinz, 2020; Burgoyne and Cain, 2022). For example, Al Otaiba et al.'s (2009) findings suggest that young children with DS (aged 1–6 years) have access to 100–200 children's books in the home and are read to daily for 10–30 min. Our findings also extended Al Otaiba et al.'s work by suggesting that mothers begin establishing these patterns when their children are toddlers. Thus, it seems that toddlers with DS, at least those in our study who came from relatively high socioeconomic backgrounds (e.g., almost all mothers had some college education), have rich HLEs that include access to books and regular shared storybook reading experiences with their caregivers.

Additionally, most mothers in our study reported using interactive reading styles (i.e., point out details from the story that are outside the written text, relate the story's content to their child's everyday experiences, teach alphabet letters and/or sounds, ask their child questions about the story and follow-up with answers) during shared book reading with their child. Our results are consistent with Barton-Husley et al. (2020) who reported that mothers of children with DS asked more questions and used more descriptions, gestures, and labels during a caregiver-child shared storybook reading activity than mothers of neurotypical children. Although the current study relied on a parent-reported questionnaire to characterize book reading styles and did not include a comparison group, our findings indicate that mothers of toddlers with DS are using interactive strategies during shared storybook reading.

Lastly, we asked caregivers to report on their child's engagement during shared storybook reading activities. Half of the mothers reported that their child asks them to read to him/her or pretends to read a book on their own during a typical week. Most mothers reported that during typical shared storybook reading activities, their child "very frequently" grabs for and/or holds the book and turns the pages. However, most mothers reported that their child was less likely to engage during the book reading activity with more advanced engagement behaviors (i.e., naming familiar pictures, asking questions about the story, or filling in words/lines of a familiar story). Our questions on child engagement were adapted from Peeters et al. (2009) who examined early patterns of child engagement (e.g., grabbing for or holding the book, turning pages of the book, pointing to pictures or words on the page) in slightly older neurotypical children who were on average 72 months old. Our pattern of results likely reflects our participants' younger ages (11–29 months) and are consistent with Barton-Husley et al. (2020), who reported that during shared storybook reading activities, toddlers and preschool-age children with DS used more gestures (e.g., pointing to pictures, head nods) and vocalizations (i.e., intentional communicative sounds) than their neurotypical peers but fewer verbalizations (i.e., words, word approximations). Additionally, Lusby and Heinz (2020), noted that when caregivers of 1-to-6-year-old children with DS were asked about child engagement during shared storybook reading activities via a caregiver-reported questionnaire, roughly 70% of children were reported to "often/always" point to pictures in the book or turn the pages during shared storybook reading activities.

4.2. Quality of a shared storybook reading task

Our second research question was to assess the quality of a shared storybook reading activity between toddlers with DS and their mothers. Our results suggest that mothers of toddlers with DS used a high percentage of expansion-comments, labels, and labeling questions during shared storybook reading. They used a moderate amount of expansion-bridges, yes-or-no questions, comments, and requests. In contrast, they used a low percentage of tag, descriptive, complex, and choice questions. This pattern of results appears consistent with previous research examining young children with DS that suggests mothers adapt their language to meet their child's developmental level (e.g., Barton-Husley et al., 2020; Burgoyne and Cain, 2022). For example, Barton-Husley et al. (2020) reported that mothers of children with DS used more questions, descriptions, gestures, and labels (i.e., interactive reading strategies) during a caregiver-child shared storybook reading activity when compared to mothers of neurotypical children who simply read the text verbatim. As children with DS get older and develop stronger language skills, mothers may begin using more complex reading strategies. This would be consistent with previous research on older neurotypical children, in which mothers used more complex reading strategies such as talking with their child about the story in ways that extend beyond the written text, expanding on their child's utterances, and asking complex, descriptive, and/or abstract questions about the story events (Altwerger et al., 1985; Haden et al., 1996).

4.3. Predicting child word learning

Our third research question asked if the richness of HLEs and the quality of caregiver-child shared storybook reading, as well as child engagement in shared storybook reading and the home language environment, related to receptive vocabulary concurrently and 6 months later. Child engagement in shared storybook reading activities emerged as the strongest, unique predictor of child receptive vocabulary concurrently and 6 months later; children who were reported as being more engaged had larger receptive vocabularies at both time points. This relation may be explained in one of three ways. First, children who have stronger receptive vocabularies may be more engaged in book reading (e.g., van der Schuit et al., 2009). Second, children who are more engaged in book reading may develop stronger receptive vocabularies because of actively participating in shared storybook reading activities. Third, there is a transactional relationship occurring between child engagement and child receptive word learning, in which children with stronger receptive language are more likely to engage in book reading activities with their caregivers. Then, as a result of spending more time with their caregiver in shared storybook reading, these children continue to develop stronger receptive vocabularies (Sameroff, 1975; Mattie and Hadley, 2021).

The home language environment, as measured by adult word count, was also a significant, unique predictor of child receptive vocabulary concurrently and 6 months later. Interestingly though, these variables were inversely related: the larger the adult word count, the smaller the child's receptive vocabulary. Barton-Husley et al. (2020) found a similar pattern when examining maternal input and child language comprehension during shared storybook reading activities in young children with DS. Those mothers of children with DS were found to use fewer total number of utterances when their child's receptive language skills were higher (Barton-Husley et al., 2020). Similarly, in the current study, caregivers of toddlers with less language talked more to their child. This may be a reflection of caregivers adapting their language to the needs/abilities of their child to help them learn new words and communicate. For example, caregivers of toddlers with higher receptive language skills may talk less because they are giving their child more time to talk (Mattie and Hadley, 2021) and/or more time to process what is being said to them. This is consistent with neurotypical literature showing that caregivers alter the amount and nature of the literacy experiences they provide based on the abilities of their child (e.g., Senechal and LeFevre, 2014).

In contrast to child engagement and the home language environment, the HLE did not emerge as a significant predictor/correlate of the child's receptive vocabulary at Time 1. Although previous research indicates rich HLEs are important to language development (e.g., Bus et al., 1995), this effect may not have been strong enough to observe in our data at Time 1, particularly in combination with our very young children with DS who were still in the early stages of language learning. At these early stages, the children's language may just not have been developed enough to capture the importance of the HLE on receptive vocabulary. However, with ongoing exposure to print-rich environments and its cumulative impact over time, in addition to the cognitive and language development of the children, the HLE did emerge as a significant predictor at Time 2. This is consistent with previous research suggesting that HLEs are associated with vocabulary and account for approximately 40% of the variance in vocabulary growth of preschool-age neurotypical children (Storch and Whitehurst, 2001).

Quality of the shared storybook reading task also did not emerge as a significant predictor/correlate of the child's receptive vocabulary at Time 1. However, it was a significant predictor of child word learning at Time 2. Similar to our findings for the contribution of the HLE on child word learning, it is possible that this effect may not have been strong enough to observe at Time 1 given our young participants and their early developmental levels. However, this effect became stronger at Time 2, perhaps because of the child's increased exposure to shared storybook reading activities over time and their increasing developmental level. This finding at Time 2 is consistent with previous research suggesting that caregiver-child shared storybook reading provides opportunities for caregivers of neurotypical children and preschool-age children with DS to scaffold their child's language based on their current language and literacy skills, therefore, enhancing their child's comprehension (Altwerger et al., 1985; Mason and Allen, 1986; Barton-Husley et al., 2020).

Two additional regressions, one predicting word learning at Time 1 and one predicting word learning at Time 2, were run with age entered as a control variable in step 1. The overall pattern of results did not change for word learning at Time 1: when age was entered alone, it significantly predicted child receptive language, but when the other variables were added to the model, child engagement and the home language environment (AWC) were the only significant predictors. The pattern of results changed slightly when predicting word learning at Time 2: when age from Time 1 was entered alone, it significantly predicted child receptive language. However, when the other Time 1 measures were added to the model, the only significant predictor was child engagement. Age, HLE, shared storybook reading (SBR), and the home language environment were no longer significant. Thus, it seems that while chronological age may be important to consider, it is not as predictive or supportive of receptive vocabulary for young children with DS as other variables, particularly child engagement and the home language environment.

4.4. Limitations and future directions

There are several limitations of this study worth noting. First, our study relied solely on a parent-reported questionnaire to measure the richness of the HLE and child engagement in shared storybook reading. This may provide less reliable and/or valid data than direct assessments/observational data, especially because participants may have responded in socially desirable ways. For example, on the HLE questionnaire, caregivers may have exaggerated their estimates of the richness of the HLE and child engagement in shared storybook reading and provided biased information, even if unintentionally. Future research could develop and work to standardize and validate measures of the HLE. It is also possible that only parents who foster rich HLEs were interested in participating in the study. However, parents of neurotypical children have been found to be accurate reporters of the HLE (Boudreau, 2005). Regardless, future research should examine the impact of child engagement in shared storybook reading on language learning in DS by directly observing the child's HLE, including caregiver-child shared storybook reading activities in the home similarly to Barton-Husley et al. (2020), who visited the participants' homes for a single 1 to 2 h time segment and video-recorded a mother–child shared storybook reading dyad to examine maternal input and its impact on receptive language outcomes. In addition, future studies should examine the impact of the HLE and child engagement on vocabulary using applied longitudinal data analysis to observe change in vocabulary growth over time.

Second, the limited number of participants in our study ($n = 13$) makes it difficult to generalize results to the larger population of children with DS. We also had more male than female participants. Further, our sample had limited diversity, including across race and ethnicity, and we did not measure if children were exposed to additional languages. Given our small sample size, we did not include socioeconomic status in our analyses, but it is important to note that most of our participants came from families with higher socioeconomic statuses (e.g., college educated mothers, most making more than $50,000 per year). Future research with larger and more diverse samples that also considers the impact of socioeconomic status is warranted. However, this study is strengthened by its use of a longitudinal design with a 6-month interval and the participant's narrow age range (11–29 months). Additionally, while the sample size was limited to 13 participants at Time 1 and 10 participants at Time 2, we found significant effects in our study, strengthening our confidence in the findings.

Third, our study did not include any comparison groups, making it difficult to know if/how our results compare to other populations, including neurotypical toddlers or toddlers with other intellectual and/or developmental disabilities. Therefore, future research should examine how HLEs of toddlers with DS compare to those of other children.

Fourth, in our study, a LENA Recorder DLP© was used to measure the quality of a caregiver-child shared storybook reading task which solely captured the audio of speakers. Therefore, we were unable to observe non-verbal behaviors of toddlers with DS which may not fully represent the child's communicative behaviors. Future research should use video recording to capture the non-verbal behaviors of children with DS and their parents during a caregiver-child shared storybook reading activity. Additionally, due to limitations with the LENA Recorder DLP© software, we were only able to measure adult word count for the entire recording period (up to 16 h while the child was wearing the device) and not the shared storybook reading activity in isolation. In addition, the LENA Recorder DLP© does not differentiate speakers, only if the speaker is an adult or child. Therefore, adult word count contains language input from any adult that is close to the child. Future research could also use audio and video recordings to code for caregiver-child interactions in the home to measure the home language environment more precisely (e.g., child-directed speech vs. total language exposure in the home). Future research should also examine the quality and complexity of caregivers' language, for example to see if it changes over time and how this relates to and/or is impacted by the child's speech and language development and to measure if there are differences in the language used by parents who talk more versus less (e.g., number of labels, questions, or comments used).

Finally, while our study, along with previous research in the field, relied solely on the role of mothers on their child's language and literacy development, future research should examine other caregivers' input on children's early development, including, for example, fathers and grandparents. Future research should also consider the impact of having multiple caregivers contributing to HLEs and shared storybook reading experiences.

4.5. Implications and application

Early childhood educators, interventionists, and other practitioners play a crucial role in educating caregivers of toddlers with DS on how to promote positive language and literacy outcomes (Fidler, 2005). Understanding the impact HLEs have on language development can help caregivers and practitioners promote a language-rich environment. These results suggest that professionals working in early intervention settings should teach caregivers of toddlers with DS practical ways to embed literacy-related activities into their child's everyday life as well as how to promote child engagement in shared storybook reading activities (e.g., holding the book, turning pages). Additionally, professionals could work with caregivers to teach what types of literacy materials are developmentally appropriate for their child and how to incorporate interactive reading strategies into shared storybook reading times to promote a literacy-rich environment. Lastly, all early childhood practitioners should actively involve caregivers in home treatment sessions and carryover programs to empower them to help their child acquire stronger communication skills.

Data availability statement

The datasets presented in this article are not readily available because they include audio files that cannot be distributed. Requests to access the datasets should be directed to LM, ljhahn@illinois.edu.

Ethics statement

The studies involving human participants were reviewed and approved by the University of Illinois at Urbana-Champaign IRB. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

Author contributions

MD, SL, and LM conceptualized the study and drafted and edited the manuscript. MD transcribed and coded the data. MD, SL, and LM assisted with data analysis. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2023.1143369/full#supplementary-material

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The co-construction of a reading assessment measure with adults with Down syndrome: a meaningful literacy approach

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Introduction: The need to develop appropriate measures of broad-based reading-related literacy skills for adults with Down syndrome has been highlighted in the literature. In this study we aimed to co-construct a valid and reliable assessment measure that can be used to document meaningful everyday reading, in adolescents and adults with Down syndrome.

Methods: The study was carried out in two stages. Stage 1 used an inclusive participatory design in which individuals with Down syndrome were research collaborators ($n=46$). Items to be included in the measure were identified and ecological, face and content validity were established through an iterative process. In stage 2 we examined the reliability of the tool and explored potential relationships between meaningful reading score and (1) age, (2) receptive vocabulary, and (3) reading ability as measured by standardized assessments. In addition, we profiled what a pilot cohort of adults with Down syndrome read ($n=33$) and how they experience reading in their everyday lives.

Results: Results showed that 46 items were generated for inclusion in the Meaningful Reading Measure (MRM). Our preliminary data showed that the tool has internal and external reliability and ecological and content validity. There were no associations between meaningful reading score and any of the other variables examined. There was considerable variability in items read (range 12–44) which reflected a broad range of reading practices. Adults with Down syndrome identified the importance of reading as a pleasurable activity and as something that aids learning.

Conclusion: The MRM developed here can be used (1) as a reading intervention outcome measure to complement existing standardized tools, (2) to profile meaningful reading in adults with Down syndrome, (3) to guide reading module content, and (4) to capture change in adults' perceptions of themselves as readers. Future work is needed to establish the tool's sensitivity to change over time.

KEYWORDS

meaningful reading, adults, Down syndrome, co-construction, assessment, literacy, participatory research

Introduction

Reading skills play an important role in the lives of people with and without Down syndrome. Reading is a key part of human communication; is required to navigate the modern technological world; facilitates participation and inclusion in society; is a recognized goal of human development; and is considered to improve an individual's overall well-being (Maddox, 2008; Dukes and Ming, 2014; McClure et al., 2015). The value of learning to read has become even more significant in recent years, as how we use written language in society has evolved. A recognition of this change is reflected in the inclusion of literacy as a global target in the UN sustainable development goals. The aim being that "by 2030 all young people and adults across the world should have achieved relevant and recognized proficiency levels in functional literacy that are equivalent to levels achieved at successful completion of basic education" (Global Campaign for Education, 2023). An increasing dependence on technology has resulted in text messaging often replacing conversation, emailing replacing phone calls and search engines replacing reference books and guides. Consequently, reading is key to navigating everyday situations and is required to function effectively both online and in the real world. In the current study, we aimed to co-construct a valid and reliable assessment checklist that can be used to document meaningful, everyday reading, in adults with Down syndrome.

The significance of reading in the everyday lives of adults with Down syndrome

The impact of how we now use written language in society and therefore the ability to read is particularly pertinent for people with Down syndrome. Firstly, the cognitive and linguistic profile of people with Down syndrome places them at risk for reading difficulties (Abbeduto et al., 2007). Many people with Down syndrome have weak phonological awareness skills (Lemons and Fuchs, 2010) and limited auditory short term memory (Jarrold and Baddeley, 2010; Godfrey and Lee, 2018), both skills that are used during phonic decoding (Cohen et al., 2008). Moreover, even those who have proficient word recognition skills find it difficult to recall details of text (Farrell and Elkins, 1994/1995). Engaging in both tasks simultaneously requires the use of working memory, an ability that is limited for those with Down syndrome (Bird et al., 2001). In addition, people with Down syndrome have language difficulties that are disproportionate to their level of intellectual disability (Abbeduto et al., 2003; Frizelle et al., 2018) and have significant difficulties understanding complex syntax (Frizelle et al., 2018). Consequently, reading comprehension is challenging and even "good readers" show a considerable discrepancy between their ability to recognize words and to understand the text (Boudreau, 2002; Byrne et al., 2002; Nash and Heath, 2011).

The second reason why changes in the use of written language in society are particularly relevant for people with Down syndrome, relates to the fact that an increasing number of people with Down syndrome are engaging independently with local communities to access education, employment and local amenities. This requires a minimum level of reading ability to allow them to complete many of the tasks associated with community living. Increased opportunities for independent living also highlight the need to develop stronger social support networks (Jobling et al., 2000) and to adopt a broader social approach to reading. This is an approach in which reading is viewed as part of living life; the social practices of reading in different contexts are considered: and the meaning that reading has in the lives of those who use it is deemed important (e.g., Papen, 2005; Taylor, 2006). Taking this approach has the potential to allow adults with Down syndrome to experience an increased connection with others and improved access to a range of social and community activities.

The need for a meaningful literacy approach

Despite the challenges experienced by people with Down syndrome when learning to read, literature shows that people with Down syndrome can and do learn to read to varying degrees (Bochner et al., 2001; Moni and Jobling, 2001; Næss et al., 2012; Reichow et al., 2019). In addition, although early onset cognitive decline is prevalent in individuals with Down syndrome (McCarron et al., 2017), cognitive development continues into adolescence and adulthood (Chapman et al., 1998). Accordingly, there is an increasing acknowledgement in the literature that reading instruction should continue beyond the years of compulsory education. Moreover, it is suggested that the young adult years may be the optimal time to focus on literacy development (Moni and Jobling, 2001) particularly when appropriate teaching and learning strategies are used (Alfassi et al., 2009; Morgan et al., 2013; Browder et al., 2014). Unfortunately, school-based conceptualizations of reading continue to dominate teaching methods, resulting in the marginalization of everyday reading practices (Katims, 2000; Maddox, 2008). Reading instruction needs to move beyond book based activities that only support the development of phonological awareness and phonic decoding skills. A purely functional approach to reading has also been criticized where only sight word instruction is used to teach words that are focused on basic survival (such as STOP or TOILET signs). Authors have highlighted that this makes up only one small part of reading and that a functional focus can often be at the expense of the development of reading for communication, education, participation and pleasure (Cologon, 2012). If programs are to be relevant for adults with Down syndrome they need to combine a functional with a social practices approach to reading (Street, 2003), to note the intention for reading, and to consider how people with Down syndrome construct reading in different contexts (Morgan et al., 2013). Instruction should also include popular culture as well as topics that are of interest and meaningful to people with Down syndrome in their everyday lives (Moni and Jobling, 2008). We refer to this as a *meaningful literacy* approach and use the term in a similar manner to Deagle and Damico (2016).

One approach in which both functional and social practices of reading are considered (*meaningful literacy*) is the Literacy and Technology Hands-On (LATCH-ON) post-school program of instruction for adolescents and adults with Down syndrome (Moni and Jobling, 2000). The program is based on the assumption that those who participate in literacy activities in their communities do so for reasons that are meaningful to them and for desired outcomes. Consequently, the program modules make explicit connections between what participants read and discuss, to events in their family and community lives.

Searching for appropriate measures of reading in the community

Although findings from the LATCH-ON program clearly indicate that adults with Down syndrome can continue to learn to read (Moni et al., 2018), gains reported have been variable and small for some participants (Moni and Jobling, 2001). In addition, the authors noted difficulties in finding appropriate measures of reading related literacy skills to measure the impact of this type of program. They highlighted that the use of measures such as the Neale Analysis of Reading Ability–Revised (Neale, 1999) or the Woodcock Reading Mastery Tests–Revised (WRMT-R) (Woodcock, 1987), could not adequately capture reading in a socio-cultural context and suggest that a broader range of qualitative measures or observations might be more appropriate. In particular, these measures do not capture change in adults perceptions of themselves as readers and are not meaningful in relation to the everyday lives of people with Down syndrome and the purpose for which they use reading.

Down Syndrome Ireland has been a provider of further education courses for people with Down syndrome in Ireland since 2012, and has provided data to the University of Queensland, Australia, to examine the longitudinal effects of the LATCH-ON program. A recognition that these standardized reading tests were inadequate for the purposes of measuring *meaningful* reading led us to search for a different measure that would allow us to document reading in everyday environments. Unable to find an existing assessment or checklist designed to explore everyday meaningful use of reading skills in this population, we chose to work together with adults with Down syndrome to co-construct an assessment checklist. A valid and reliable checklist could be used as a baseline and outcome measure for post-school reading programs and to guide the content of modules to ensure that programs are meaningful to everyday social practices and reflect participants' own preferences/choices. This type of checklist may be more sensitive to change in relation to the range of items read pre- and post-intervention programs and could also complement standardized tools that are more focused on more specific skills, such as decoding.

Psychometric properties of an assessment tool

To use an assessment checklist in clinical practice or for research purposes, it must have evidence of sound psychometric properties (Andersson, 2005; Dockrell and Marshall, 2015). These include the overarching concepts of validity, reliability and responsiveness (Mokkink et al., 2010). Reliability is an indication of whether the measurement tool gives consistent results each time it is used. One indicator of reliability is *internal consistency* which is an index of how far the different items that make up a scale are measuring a common construct or idea. A second reliability measure is referred to as *external reliability* and is a measure of how consistent the scores are between two or more test sessions taken in close proximity (test–retest reliability). In contrast, validity indicates how well a test captures what it sets out to assess. It is measured in a number of ways and includes concepts such *as ecological, face, content,* and *concurrent validity*. *Ecological validity* examines whether the items included in the test are reflective of those in real life settings. *Face validity* refers to whether the test appears to assess the target reading practices in question and includes aspects such as the overall appearance of the test and how it is presented. *Content validity* is the degree to which the content of the test is an adequate reflection of the construct being measured. For example, the content should reflect a wide range of items so that respondents have the opportunity to portray the extent of their skills in a given area. *Concurrent validity* refers to how well the scores on a new measure correspond to those on well established "gold standard" tests for the same children. Note, we do not include all measures of validity in this study as *predictive validity* for example is more appropriate to child populations where significant development is expected regardless of any intervention input. In addition we have not measured *responsiveness* (the ability to detect change over time in the construct being measured) as participants were engaged in different programs over varying time frames and it was not possible to use the tool to measure their skills at baseline.

Current study

This study was carried out in two stages. In stage one we used an inclusive participatory research design in which adolescents and adults with Down syndrome were partners in the research. This design is collaborative such that (1) the research is undertaken *with* rather than *about* people with Down syndrome (Walmsley, 2004) and (2) their input / opinions and perspectives are integral to the work carried out. The adults with Down syndrome who worked with us in stage 1 are referred to throughout as our *collaborators*. In stage 2 we have taken a more traditional approach and refer to this cohort as *participants* throughout the study.

The following research questions are addressed:

Stage 1

- What items should be included in an assessment tool that can be used to profile meaningful reading in adolescents and adults with Down syndrome?
- How should the tool be presented so that (1) it is accessible for people with Down syndrome (2) it has acceptable face and content validity?

Stage 2

- To what degree is the Meaningful Reading Measure (MRM) developed reliable?
- Is there a relationship between meaningful reading scores and (1) age, (2) receptive vocabulary, and (3) reading ability as measured by standardized assessments.
- What do adults and adolescents read in their day to day lives?
- How is meaningful reading experienced by adults and adolescents with Down syndrome in relation to what, when, where and why they read; their preferred medium; what is hard about reading; the best thing about reading; and what they would like to be able to do with their reading?

With reference to measuring concurrent validity usual practice is to examine the relationship between the new measure and the "gold standard" currently in use (with the expectation that a relationship will exist) and the investigation is about determining the strength of that relationship. Where a gold standard does not exist, the investigation is one of determining if any relationship exists (hypothesis testing). Given that there is no recognized reference standard for measuring meaningful reading in adults with Down syndrome, our comparisons with standardized tests were therefore considered to be hypothesis testing (rather than measures of concurrent validity). This is in keeping with the Consensus Based Standards for the Selection of Health Status Measurement Instruments (COSMIN) taxonomy[1] (Mokkink et al., 2010). We did not anticipate a relationship between receptive vocabulary and meaningful reading score. Vocabulary assessments are developed so that the test items increase in difficulty/abstractness as one progresses through the test. In contrast, our meaningful reading checklist is designed to reflect a broad range of items in a real-world context, rather than items that increase in difficulty. Similarly, we did not anticipate a relationship between our standardized reading checklist and meaningful reading score. This hypothesis was based on the fact that our tool was developed to capture changes in the range and number of items that people with Down syndrome may read in a socio-cultural context, a need that is currently not met by standardized reading measures (Moni and Jobling, 2001). Given (1) that reading underpins both our meaningful measure and standardized measures and (2) the relationship reported between vocabulary knowledge and word identification abilities (Wise et al., 2007) one might argue that a relationship is possible, although we did not anticipate relationships would exist. Therefore, we needed to examine this empirically to be sure if our hypotheses were correct.

With respect to potential relationships between meaningful reading score and age, we could have hypothesized a negative association, as younger adults with Down syndrome are less likely to experience cognitive decline than those who are older (McCarron et al., 2017). In addition, they are more likely to have been educated in a mainstream school, and therefore have better reading outcomes (de Graaf et al., 2013). On the contrary we could have anticipated a positive association with age, as older adults have greater life experience and are therefore more likely to be engaged independently with education, employment, or their local community for a longer period, giving them greater exposure to the type of items that would be included in the checklist.

Methods

Ethics statement

Ethical approval was granted from the Clinical Therapies Social Research Ethics Committee at University College Cork.

Stage 1—creation of assessment tool

Research partners/collaborators

Forty-six adults with Down syndrome were recruited in to stage 1 of the study. Adults were recruited through three adult education classes, two delivered online and one in-person, all administered through Down syndrome Ireland (DSI). DSI is an organization which offers support and services for people with Down syndrome and their families in Ireland. The final author facilitated study recruitment by liaising with the adult education course teachers. The education courses were offered to all adult members of DSI in 2021, regardless of reading ability. Down syndrome Ireland take a universal design approach to learning and therefore there are no literacy, academic or social pre-requisites to participating in the classes. Online classes required access to the internet and an online platform. The aims of the classes were to build friendships and to develop literacy skills in a very broad sense. All collaborators spoke English as the primary language of the home and based on teacher report, were of mixed cognitive ability. We did not assess the overall cognitive ability of collaborators as we deemed this to be inappropriate in the context of developing a meaningful reading tool with adults and were not using IQ as an inclusion/ exclusion criterion. This is in keeping with Greenspan and Woods (2014) who suggest that arbitrary IQ test scores provide little insight into the relative cognitive strengths and weaknesses of adults with ID. Demographic information for collaborators from each of the classes is given in Table 1.

Procedure

Creating the tool

Collaborators attending each of the three classes were given an easy read information sheet with visual supports explaining the purpose of the study and what would be involved. The information sheet was also read orally by the course teacher who explained that those who wished to be included would complete an optional additional literacy exercise. Collaborators were also invited to ask the researchers, their teachers or parents for further information if needed. This is in line with the Health Service Executive (2022), to maximize a person's capacity to consent through supported decision making. Collaborators were provided with a written consent form, requiring a tick box response, or could choose to give consent verbally. The consent form was modeled on a previous form which was co-designed with the Down Syndrome National Advisory Council (a committee of people with Down syndrome). The first stage of the literacy exercise was to notice and write down all the things the participants read in a week. The task was outlined on the first teaching day of the week (Monday for the online courses) and a discussion took place in the class or online about potential items that participants might read each day. Results were gathered within 1 week of the initial discussion. Three students were absent when the majority of the class completed the task and they elected to complete it on their return.

All anonymized results were shared with the researchers who compiled the lists into one draft checklist and created the first version of the Meaningful Reading Measure (MRM). Guidelines on easy-read materials from Nomura et al. (2010) and the UK Department of Health (2010) were considered in the design of all aspects of the tool. Questions were concise with limited use of abstract language and picture supports were provided to support the meaning of each question. The layout contained wide margins with consistent spacing. Most text was placed inside a defined space in a clear non-serif font and a type-size of 18 pt. or greater. In addition, key words were boldened for emphasis (see Figure 1).

Version 1 of the MRM contained three sections. Section 1 focused on demographic information, including age; whether collaborators were completing the checklist independently or with support; and educational information, such as number of years spent in mainstream/special education and any additional courses completed (Figure 1A). Section 2 included a list of all items that collaborators stated they read. Items that were very similar were conflated (e.g., text

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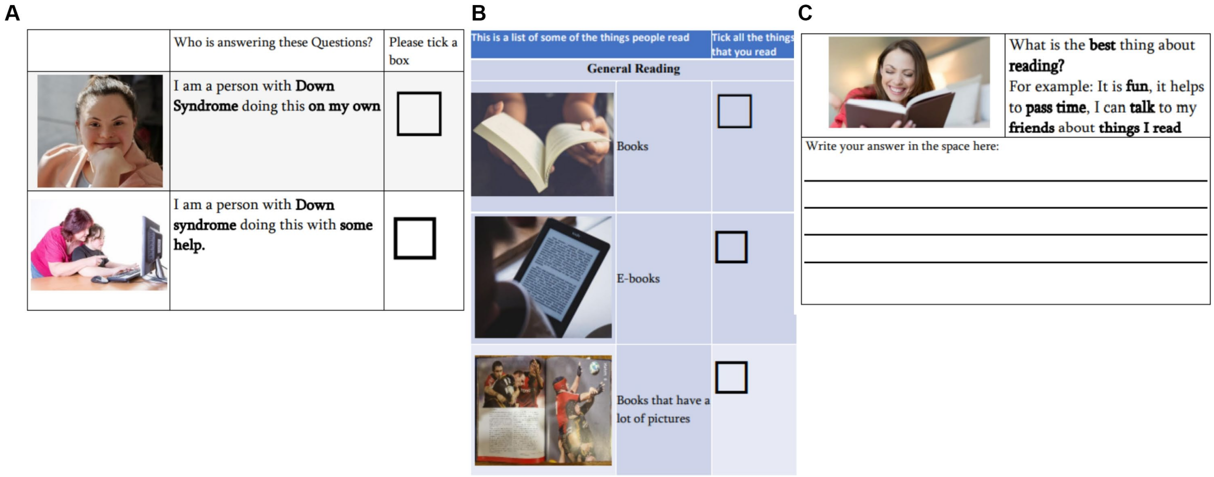
[1] http://www.cosmin.nl

TABLE 1 Collaborator demographics for checklist creation group *n*=46.

Class	Age range			Male		Female	
	(Years)	n	%	n	%	n	%
Online adult cohort	18–29	24	52.2	12	26.1	12	26.1
Online teenage cohort	16–18	13	28.3	5	10.9	8	17.4
In-Person cohort	20–55	9	19.5	3	6.5	6	13
Total by gender		46	100	20	43.5	26	56.5



Who is answering these Questions?

Please tick a box

I am a person with **Down Syndrome** doing this **on my own** ☐

I am a person with **Down syndrome** doing this with **some help.** ☐

This is a list of some of the things people read

Tick all the things that you read

General Reading

Books ☐

E-books ☐

Books that have a lot of pictures ☐

What is the **best** thing about **reading?**

For example: It is **fun**, it helps to **pass time**, I can **talk** to my **friends** about **things I read**

Write your answer in the space here:

FIGURE 1
Sample image from version one of the meaningful reading measure.

message, what's app message) resulting in 42 unique items. Each item was depicted visually with supported text and a tick box which would allow collaborators to indicate which items they typically read. Each ticked item would be counted as 1 which when summed would result in a total meaningful reading score. Space was also allowed for participants to add items which were not covered in the checklist. Figure 1B shows an example of how the items were depicted. Section 3 included 7 additional questions on collaborators' reading, to provide a context for collaborators' responses and to inform future courses where meaningful reading is an area of focus. The first was a closed question about the medium through which collaborators read and why, with a number of options *Do you most like to read—a paper book, an eBook, on a tablet or computer, listening to audiobooks, I do not like to read.* The remaining questions were open-ended: *why do you read?; where do you read?; when do you read?; what is the best thing about reading?; what would you like to be able to do with your reading?; and what is hard about your reading?* (Figure 1C).

Establishing face and content validity

To establish face and content validity the MRM was then shared in electronic and hard copy formats with the 10 collaborators who were completing their adult education course in person (a sub-group of the original 46). Those who attended the online adult education classes were not included at this stage as they had completed the classes and moved on to other things. The process of obtaining feedback was iterative. The third author sent the checklist to the class teacher and asked him to get the participants' views on the MRM. An in-class discussion took place and the teacher reported back verbally to say that the collaborators liked all aspects of the MRM. We reflected on this process and realized that a request for general feedback was not an adequate method to generate feedback on specific elements of the tool. Consequently, we revised our methodology. Regarding the main section of the MRM (the list of items), we identified 15 image supports from the checklist which we deemed to be either (a) visually unclear, (b) potentially problematic for someone with visual difficulties, or (c) a poor representation of the concept the image was intended to represent. Additional images were sourced for each of these 15 items, to ascertain which image depicted the concept most clearly. These images formed the basis of a PowerPoint presentation that was given to the 10 participants with Down syndrome through the online conferencing platform Zoom. The online conference call was conducted by 3 members of the research team and in addition to the collaborators with Down syndrome, was attended by the class teacher and two teaching assistants. The class teacher acted as a facilitator for any collaborator who had difficulty understanding any aspect of the process. The PowerPoint presentation was given by the second author and consisted of 15 slides each containing 3 different images to represent the same concept. Figure 2 shows an example with reference to the item *Maps*. Collaborators were then asked which picture depicted each item most accurately/clearly. Responses were tallied and the image for which the majority of the group voted was chosen to represent each item in the checklist.

On the basis of collaborators' responses a second version of the MRM was created to include the validated image choices and a

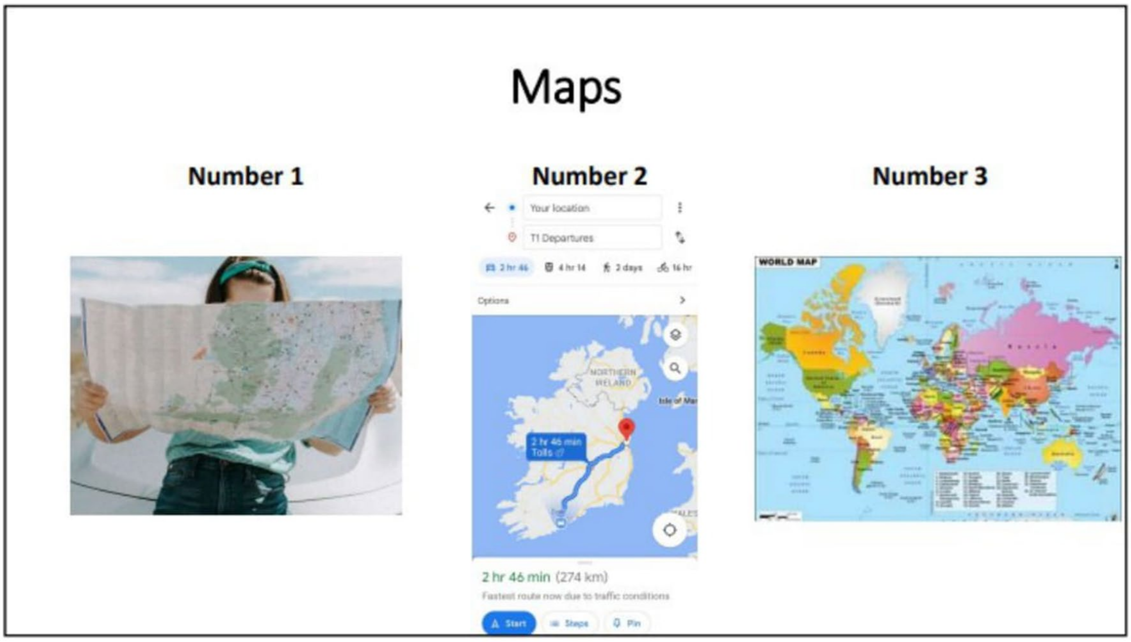


FIGURE 2
Sample from image validation exercise.

reorganization of the items such that they were presented in the following categories—general reading; personal reading; reading when out and about; and reading a screen/reading for fun. The MRM was sent to the same 10 collaborators so that they could complete the second iteration of the checklist as an in-class exercise, facilitated by their class teacher. Prior to completing the MRM collaborators were asked to appraise the measure with reference to (1) ease of completion, (2) items included, and (3) layout/overall appearance. Immediately following completion, a class discussion took place where collaborators discussed what they liked and did not like about the measure, as well as any further modifications that should be made. The in-class exercise was followed by an online focus group in which the researchers asked for final feedback on the measure. Again the class teacher acted as a facilitator, rephrasing questions when required and asking specific collaborators for their input. The focus group was audio recorded and transcribed by the second author (SOD). SOD then coded responses with one of three codes. The code *affirmation* was used to indicate a positive comment about an element of the measure; *alteration* was used to indicate a recommendation to change or remove an element; and *additional information* was used to reflect comments pertaining to the length of the survey, ease of completion and general feedback. Suggested modifications were taken into account and the final version of the MRM for teenagers and adults with Down syndrome was created.

Results

Stage 1

Version 1

In addressing our first research question we aimed to establish what items should be included in an assessment measure that could be used to profile functional reading in teenagers and adults with Down syndrome and how that measure should be presented (i.e., the ecological, face and content validity of the measure). Responses from 46 collaborators indicated that 42 items should be included in the first version of the checklist. A frequency analysis of the items listed was conducted and is shown in Figure 3. Following a review by the research team an additional 4 items were added—books with picture supports; museum displays; contracts; and board games. This resulted in a total of 46 items in the first version of the MRM.

Face and ecological and content validity

Results from the image validation process indicated that only two of the previously used images were chosen by adults with Down syndrome to remain in the MRM, namely those that represented *emails* and *board games*. The group voted on an alternative image for the remaining 13 representations. For each image there was a majority vote. The images presented through PowerPoint along with the voting tally are shown in Supplementary Figure S1.

Results from the focus group affirmed the following features of the measure—the use of visual images and supports (such as bolded text) with all questions rather than confining these supports to the checklist part of the measure; the addition of examples for sections 1 and 3 (the demographic and opinion questions); the use of a blue background color to enhance the checklist portion of the tool; and the use of sub-headings to reflect different categories of reading within the checklist. In relation to alterations, collaborators suggested increasing the font size throughout the measure; increasing the space provided for collaborator responses; the inclusion of lines to assist with response presentation; and the addition of two DSI further education course options, which had been completed by most of the collaborators. Finally, “additional information” that was noted in the focus group related to guidance on how the tool might be completed in the future. The majority of collaborators (70%) stated that their preference would be to complete the measure using pen and paper rather than using a computer. Seventy

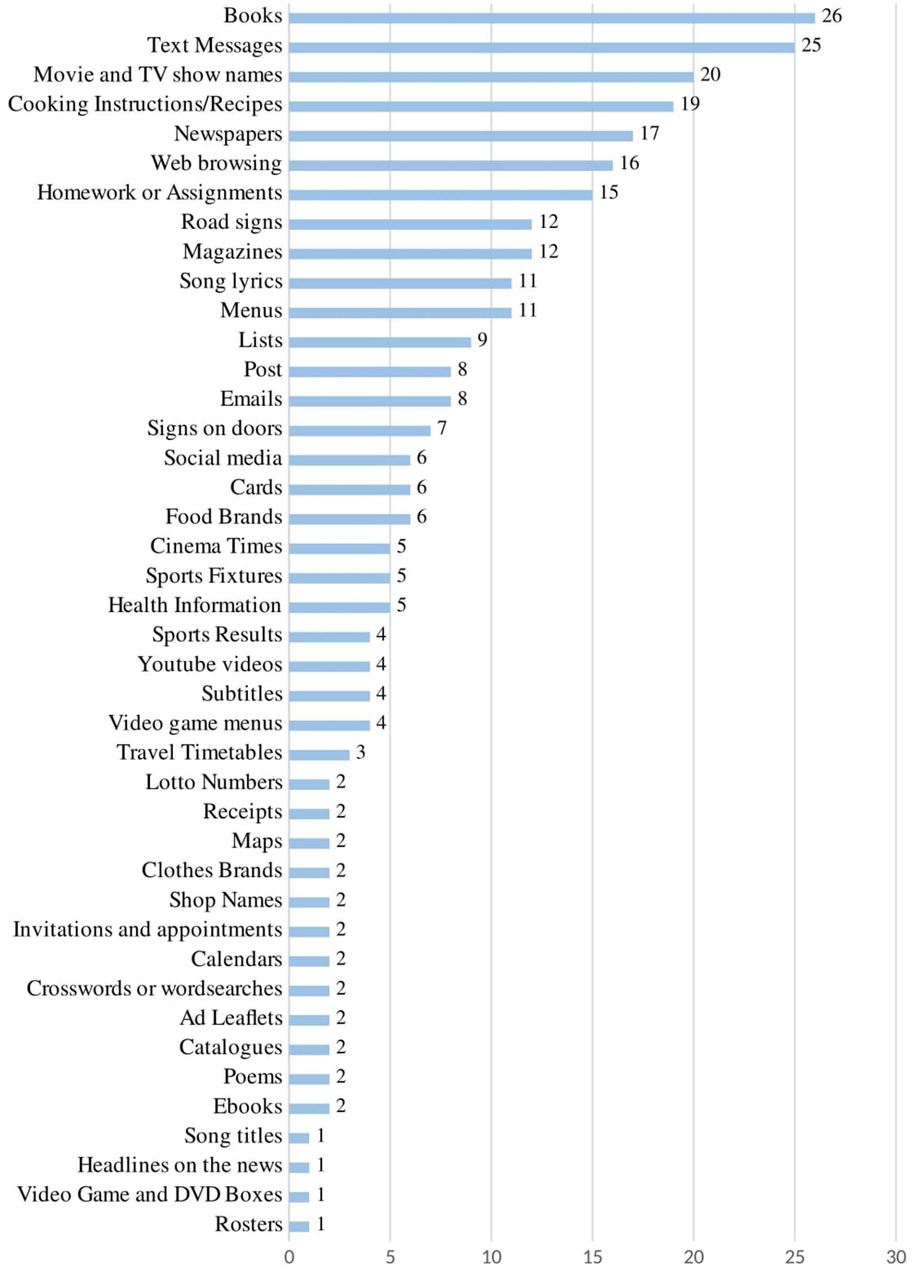


FIGURE 3
Frequency of reading items listed by collaborators.

percent also stated that they would prefer to complete the measure as part of a class exercise with the assistance of their teacher, rather than independently at home or with the assistance of a family member. All recommended alterations were made to the measure before progressing to stage 2 of the study.

Stage 2

Stage 2 included two parts. The first examined test–retest reliability and associations with standardized tests and the second examined the lived experiences of adults with Down syndrome.

Participants

In order to reduce the burden on any one group, different cohorts were recruited into the second stage of the study. To establish external reliability 23 adults with DS were recruited into the test re-test component of stage 2. These were a convenience group of adults who were studying on adult education courses run or supported by DSI and all spoke English as the primary language of the home. Participants were recruited through the third and final authors who contacted the course teachers, informed them of the study, and asked them to impart the information to those attending their course. As before, the course teacher shared the study information sheet and consent form (developed in an easy read format) with the course participants and read the information aloud, to ensure informed consent. Of the 23 that gave initial consent, 3 were absent on the day of MRM completion and 1 opted not to complete the MRM the second time. Therefore 19 adults with Down syndrome participated in this aspect of the study.

Twenty-five adults with DS were recruited into the next part of stage 2—exploring potential relationships between the MRM, age and standardized test results. These participants were a cohort for whom standardized literacy and vocabulary assessments had been recently completed as baseline and outcome measures for their literacy program. The participants were either attending an adult literacy course administered by DSI or were in the process of enrolling in one. The recruitment process was similar to that outlined above. Those in the process of enrolling in an adult literacy course were contacted directly by the 3rd author and invited to take part. The 3rd author read the information sheet and consent form to the potential participants and was available to ask any questions. Initially all of those invited elected to take part in this aspect of the research, however 1 changed her mind, 1 was absent on the day the checklists were completed and PPVT scores were not available for 2 participants. Consequently, the final number included was 23 for both reading measures and 21 for the vocabulary measure. There was an overlap of 12 participants between the test re-test group and those who completed the standardized assessments.

Thirty three participants were recruited to complete the final part of stage 2 (i.e., what adults and adolescents read and how they experience reading in their day to day lives). These included all of those who completed the test retest; those who did the standardized assessments; and 2 additional participants who were absent on the day the MRM retest was completed. For those who completed it twice, we report the MRM results from the first timepoint. Demographic information for each cohorts is given in Table 2.

Measures

Peabody picture vocabulary test

The Peabody Picture Vocabulary test-4th Edition (PPVT-4) (Dunn and Dunn, 2007) was used to measure receptive vocabulary. This is a norm-referenced standardized test which can be used from 2½ to 90 years. It has test–retest reliability coefficients of 0.92 and 0.96 and split half reliability of 0.94 and 0.95. Standard scores are based on a typically developing population and are therefore not suitable for people with intellectual disability. Consequently, raw scores were used in the current study. Participants are shown four color pictures on each page. The test administrator says a word that describes one of the pictures and the participant is asked to indicate which one of the four pictures is being described.

Burt reading test

The Burt Reading Recognition test (Gilmore et al., 1981) was used to measure participants' ability to read single words. The test consists of a list of 110 real words arranged in groups of 10, presented in decreasing size and increasing order of difficulty. The test was developed for use with typically developing children up to 12 years old. It is recommended to stop testing following 10 consecutive errors. The total number of words read correctly yields a raw score and this can be converted into a reading age. We did not deem reading age to be an appropriate metric for adults with intellectual disability and have used raw scores in this study.

Procedure

Participants completed the MRM in hard copy in the room in which they attended their adult literacy course or in the center in which their course would be delivered. Those already attending a course completed it in one sitting at the same time as their peers. Those enrolled in a course came to the education center individually and completed it while the 3rd author was present. Teacher or researcher support was available in both contexts and was dependent on individual preferences. Some adults chose to sit one to one with a teacher or teaching assistant, while others completed the task in small groups (no greater than 4 adults to one teacher providing support). In the group context teachers introduced the task to the whole group and some adults initially expressed doubts about their ability to read. In response, the teachers explained that the MRM was not just about reading books but was developed to document all types of reading that might occur in the participants' daily lives. Support given also involved reminding participants to look at each item; to turn the page; and to ask if they needed a break. Teachers also gave positive feedback and encouraged participants to get to the end of the task. Those who completed the measure twice did so within 1 week. Alpha numeric codes were added to participant response sheets so that meaningful reading scores could be cross-referenced (a) time 1 with time 2 and (b) with vocabulary and reading test results. All data were anonymized within DSI before sharing with the first and second authors.

Data analysis

Internal consistency was measured using Cronbach's alpha. Test–retest (external) reliability was initially examined using a Spearman correlation coefficient for paired samples. However, because the correlation measures only the strength of the

TABLE 2 Participant demographics for stage 2.

	Test–retest group (*n*=19)				Standardized assessment group (*n*=24)			
	M	*SD*	Median	Range	*M*	*SD*	Median	Range
Age	30	8.67	29	21–53	30.58	8.81	28.5	19–53
Sex (M:F)	10:9				15:9			
PVVT (*n* = 22)					92.32	33.74	80.5	47–257
BURT (*n* = 24)					29.13	5.97	26.5	0–96

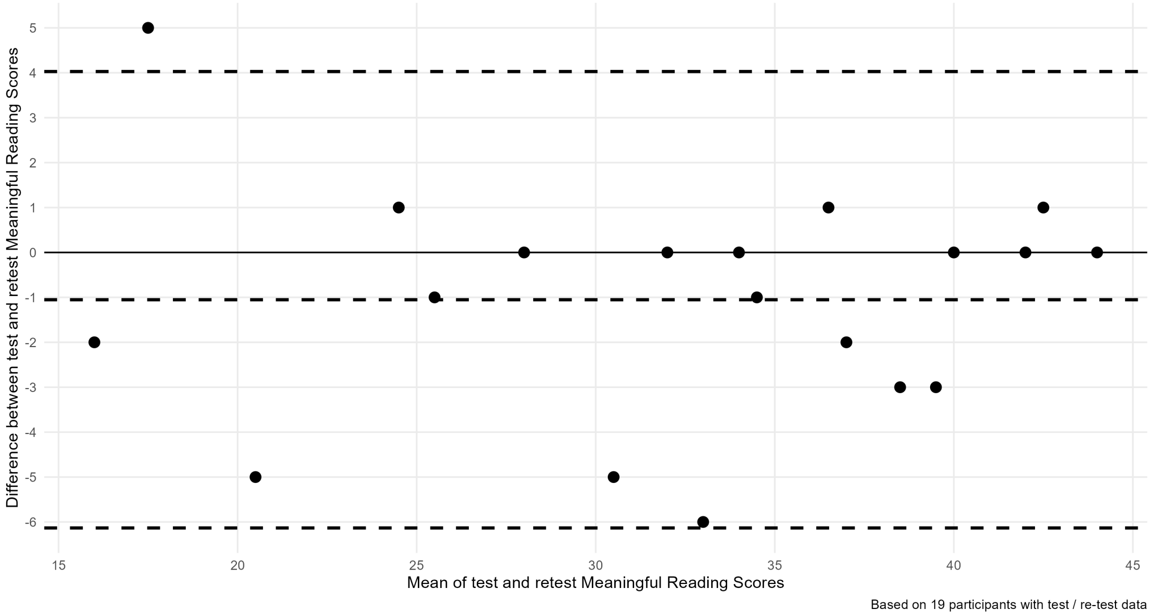


FIGURE 4
Bland–Altman plot for agreement in test and retest meaningful reading scores.

relationship between the two variables, but not the agreement, we also completed a Bland–Altman analysis (Bland and Altman, 1995) and calculated a Coefficient of Repeatability. In a Bland–Altman plot (Figure 4) the difference between the test–retest scores is plotted against the mean of the scores for each time point. This method allows us to calculate the mean difference between the two times the assessment was completed (the "bias") and 95% limits of agreement of the mean difference (1.96 SD).

Spearman correlations were carried out to examine if there were relationships between total meaningful reading score (based on the first completion of the measure) and results from standardized vocabulary and literacy assessments. Participants' raw scores were used for the standardized vocabulary and literacy assessments as these assessments have not been standardized on populations with intellectual disability, to minimize potential floor effects, and to avoid obscuring individual differences by utilizing standard scores (e.g., Kover and Atwood, 2013). Quantitative data on demographics, and meaningful reading profiles were analyzed descriptively. Finally, a qualitative directed content analysis (Hsieh and Shannon, 2005) was completed on the data collected from section 3 of the measure (i.e., responses to the open-ended questions that would allow us to profile in more detail participants' meaningful reading practices). All responses were transferred into NVivo in preparation for analysis. Steps taken were those outlined by Hsieh and Shannon (2005). Data were read repeatedly by the third author to achieve immersion and a sense of the whole data set. Data were then read word by word, and words from the text that captured key predetermined concepts (e.g., where, when, and why do you read) were highlighted to derive codes. Codes were then organized into categories based on how different codes were related. Incidence of codes representing each category were noted under each of the six question headings. To increase trustworthiness all data and coding was discussed with and reviewed by the first author.

Results

Our first research question in stage 2, was to establish to what degree the MRM was reliable (i.e., what is the internal and external reliability of the measure?). Of the 33 participants 31 had a complete data set. Cronbach's alpha (Kuder–Richardson formula-20, used for dichotomous scores) showed an internal consistency value of 0.93, indicating a homogenous test. We examined test–retest reliability of scores based on our sample of 19 participants. The estimated paired sample Spearman correlation was high at 0.95 (95% CI 0.88–0.98), while the ICC estimated with a linear mixed effects model was 0.96. We also evaluated agreement in test re-test scores using a Bland–Altman analysis (Bland and Altman, 1995). The mean difference was −1.05 and the 95% limits of agreement were −6.13 to 4.03. Visual inspection of the Bland–Altman plot did not reveal any concerning patterns or trends (see Figure 4). Finally (based on a within subjects SD of 1.93), the Coefficient of Repeatability showed that the difference between 2 observations for the same person is estimated at <5.35 points for 95% of observed pairs.

Our second research question addressed whether there was a relationship between meaningful reading and (1) age, (2) receptive vocabulary, and (3) reading ability as measured by a standardized vocabulary and reading assessment, respectively. Given that the MRM data were not normally distributed, Spearman's rank correlations were completed. Results indicated no significant relationships between meaningful reading score and age ($r = 0.12$, $p = 0.57$); receptive vocabulary ($r = 0.01$, $p = 0.96$); or reading ability as measured by the Burt word recognition test ($r = -0.05$, $p = 0.79$).

Our third research question asked what adults and adolescents read in their day to day lives. Results for the mean number of total items read (Meaningful Reading Score) are given in Table 3 and the frequency with which each item was ticked is shown in Figure 3. The lowest score was 12 and the highest was 44, indicating that everyone who participated read a minimum of 12 items on the checklist in their everyday lives. As shown in Figure 5 almost all participants (≥91%) ticked that they read *post*, (e.g., *letters/ postcards, items received by mail*), *food brands*, *shop names*, and *cards*. Approximately 50% of participants stated that they read *lottery numbers, headlines on the news* and *newspapers*. *E-Books* were the least common item read (33% of participants).

TABLE 3 Checklist results for complete cohort.

	Average reading checklist score (*n*=31)			
	M	*SD*	Median	Range
Reading score	31.06	9.49	34	12–44
Age	28.94	8.41	26	19–53
Sex (M:F)	21:12			

Our final research question addressed how meaningful reading is experienced by adults with Down syndrome in relation to when, where and why they read; their preferred medium; what is hard about reading; the best thing about reading; and what they would like to be able to do with their reading? Our qualitative analysis of these questions (asked in the final section of the meaningful reading checklist) is summarized in Figure 6.

The first question was a closed question (using a tick-box format) and asked participants about the medium in which they preferred to read and why (*Which do you most like to read? Why?*). Most participants chose to read on a tablet or computer (*n*=12); some preferred a paper book (*n*=7); others stated that they do not like to read (*n*=7); some did not have any preference (*n*=4); one participant preferred listening to audio books and no-one reported reading eBooks or using a kindle. The remaining questions were all open-ended, the first of which asked participants what they considered to be the best thing about reading. The most common

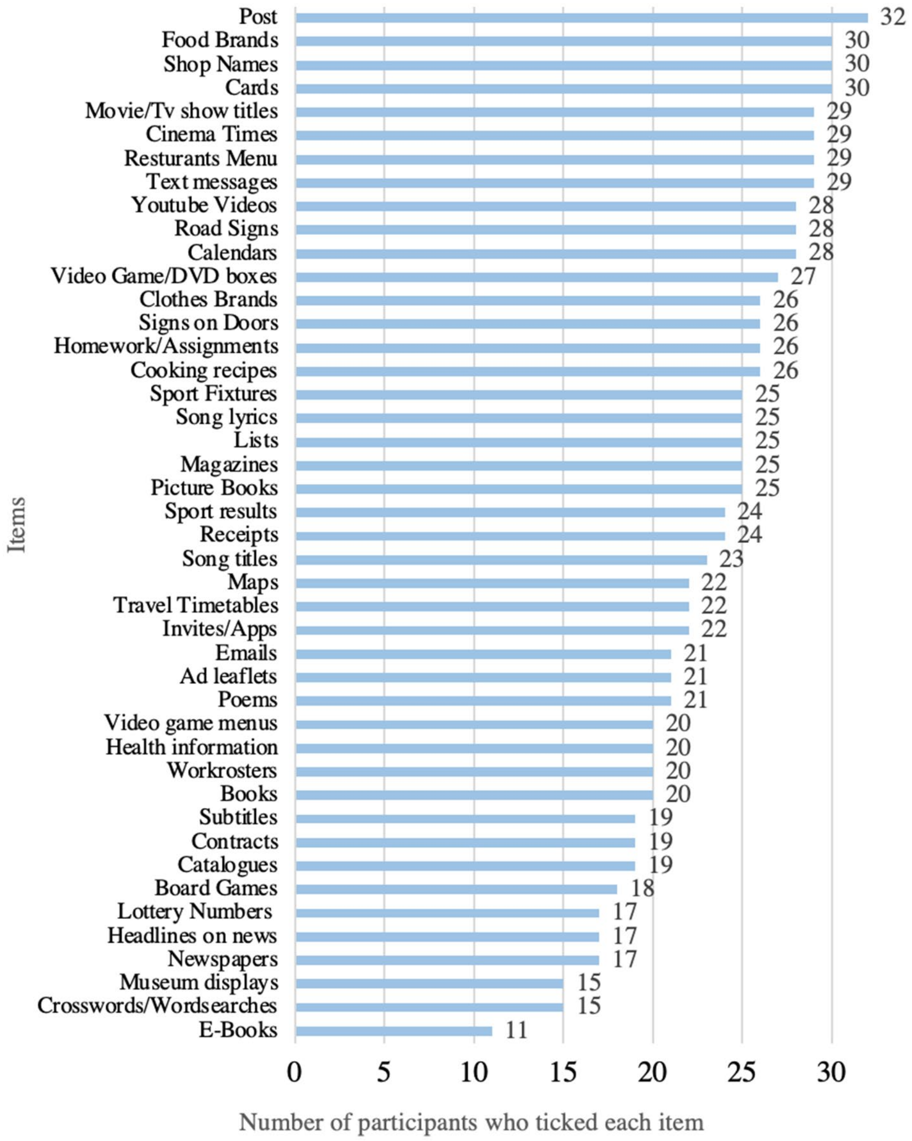


FIGURE 5
Frequency with which each item was read from stage 2 participants (*n*=31). This graph does not present data on 2 items (social media and web browsing) which were accidently excluded from the checklist presented to these participants.

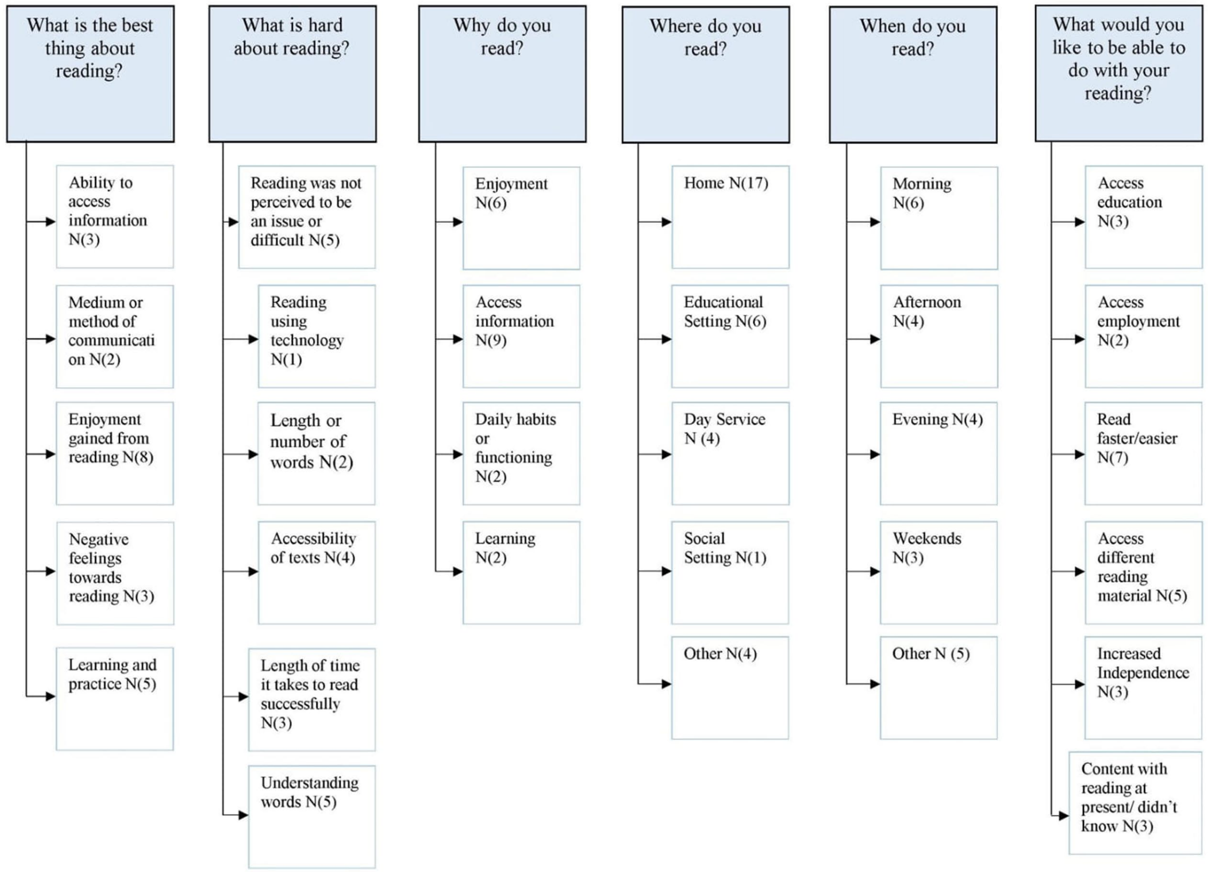


FIGURE 6
Content analysis of questions in section 3 of the assessment checklist. N=number of participants in each category.

response was that reading was pleasurable/enjoyable. One participant described the immersive nature of reading "I can read books; it is fun and is [like] a TV in my head" while another enjoyed it as a relaxing activity "just relaxing my brain." Some participants ($n=3$) described how reading enabled them to access information or find out more about their interests, "[it] gives me information." Others ($n=5$) felt that reading was helpful for learning and a way to practice getting better at other activities "Helps me to focus and learn big, long words, helps me with my phone." A couple of participants ($n=2$) stated how reading helped them communicate with others, and others ($n=3$) expressed negative feelings toward reading, "I was better years ago." In the next question participants were asked what they felt was hard about reading. The two most common responses to this question were that reading is not difficult ($n=5$), "I do not think it's hard" and that understanding words is difficult ($n=5$), "words I do not understand….it takes me *for* months to read one story." Participants also had issues with small print and with reading long words. When asked why they read, most said that they read to access information ($n=9$), "I read to find out SOAP spoilers" or "To learn new things… to find out information." Others stated that they read for enjoyment ($n=6$); because it was a daily habit ($n=2$); and to aid their learning ($n=2$), "to learn new ways to spell, syllables." In the next question participants were asked where they read. Most stated that they read at home ($n=17$); others responded that they read in an educational setting ($n=6$), others read in their day service ($n=4$), and one participant reported reading in a social setting. Other respondents said that they read only when they had to, "Only when I have to…, texts on my phone when they come in." Participants were then asked when they read. Responses were varied and included during the morning, afternoon, evening, and weekends. Five participants did not specify a particular time of the day and stated, "I do not have time to read" and "Quiet time with my mum…. midday prayers." In the final question, participants were asked what they would like to be able to do with their reading. The most common response was that participants would like to be able to read faster and more easily ($n=7$); some wanted to access other material ($n=5$) "I would like to read on my phone"; others wanted to use reading to help them achieve greater levels of independence ($n=3$) "Put money in my [my bank account] my boss put in" and "[Read] on my phone and computer and without help"; some participants did not have any goals for their reading ($n=3$); and a few would like their reading to help then access employment ($n=2$) and further education ($n=3$). More detailed quotes under each category are given in Supplementary Tables S1, S2.

Discussion

Stage 1

In the current study we initially aimed to establish (1) what items should be included in an assessment measure that could be used to profile meaningful reading in adolescents and adults with Down syndrome and (2) optimal presentation to ensure the measure has acceptable face and content validity and is accessible for people with Down syndrome. Ultimately, this type of assessment tool could be used as an outcome measure for post-school reading programs and could serve to complement standardized tools as it would potentially be more sensitive to change, regarding the range of items read pre- and post-intervention. We argue that an increase in range of items read represents one aspect of reading growth, which in the longer term could result in the ability to read a greater number of words. Importantly, an increase in range of items read has the potential to increase confidence and motivation when engaging with written material. In addition, it can facilitate individuals with Down syndrome to participate in richer social practices in their community, much of which can be achieved through engagement with a broader range of written material (e.g., through social media), without increasing the difficulty level of words presented. Additionally, the MRM could be used as a measure of cognitive decline. In practice, in early stages of dementia, parents often report a reduction in their son's/daughter's ability to engage with meaningful reading tasks that are part of their daily lives, for example finding the television channel they want. One would expect that these changes are more likely to be reflected in a meaningful reading measure than in standardized reading assessments. However, further work would need to be carried out longitudinally to examine sensitivity of the task to change over time.

Stage 1 of our study was participatory in that adolescents and adults with Down syndrome were active collaborators/research partners throughout the process of constructing the tool. Ninety-one percent of the items included in the final version of the tool were generated by 46 individuals with Down syndrome, with the remaining 9% (4 items) added by the research team. Following the generation of the items, our method of collaborative practice in relation to face and content validity was problematic, in that when asked in a general way, all collaborators with Down syndrome stated that they liked all aspects of the measure. This was a reminder to the research team that we needed to be much more specific in our approach. We then focused on how each item was visually depicted and identified 15 image supports that we deemed to be visually unclear; potentially problematic for someone with visual difficulties; or that could be represented in multiple ways. Alternative images were sourced for each of these items and collaborators were asked to vote on their preferred representation, by raising their hand. Using this method gave our collaborators a specific area to critique and allowed all members of the group to actively contribute, independent of reading or spoken language ability. Consequently, 13 of the 15 images were changed from how they were originally depicted, indicating that the images we had originally chosen were not optimal for our collaborators with Down syndrome. This is keeping with that reported by Sutherland and Isherwood (2016) who found that although photographs/images are helpful, they can often be confusing and do not always convey the correct message. Following this process, our collaborators indicated that they really enjoyed the exercise and that they had never been asked to contribute to research like this before. The realization that the images would be changed based on their opinions appeared to increase their confidence and level of involvement in the final feedback stage. Final feedback was informed by a class discussion and an online focus group, in which our collaborators were asked to focus on specific aspects of the measure, such as how easy it was to complete, the items included and the layout/overall appearance. In this stage of the process collaborators were increasingly vocal, were very forthcoming about specific aspects of the measure that they liked and gave several suggestions regarding how the presentation of the measure could be improved. Suggestions, which included increasing the font size and space provided for participant responses, were reflective of the guidelines put forward by Nomura et al. (2010) who highlight the range of layout interventions required to make a document easier to read and comprehend. It was interesting that most collaborators stated that their preference would be to complete the tool using pen and paper (rather than on a computer). This is perhaps unsurprising, given some of the ongoing computer usability challenges evinced by individuals with Down syndrome (e.g., password usability) (Kumin et al., 2012), as well as variability regarding formal computer training among our collaborators. In addition, they stated that they would like to do it as part of a class exercise with the assistance of an adult educator, rather than at home. However, both responses may just reflect our collaborators' experiences in this study and if given the opportunity to complete the tool online or at home, it is possible that they may equally embrace this experience.

Stage 2

Reliability

In the second stage of the study, we aimed to establish if the measure was reliable. Our findings clearly indicate both internal and external reliability. Internal reliability is shown by a Cronbach's alpha of 0.93, which is established in the literature as an indicator of strong reliability (Taber, 2018). In addition, although there were some individual differences in our test–retest data, a Spearman correlation of 0.95 indicates a strong relationship between the first and second time the tool was completed (external reliability). This was further supported by the agreement levels shown in our Bland–Altman analysis (mean difference of −1.05), an ICC of 0.96 and a small coefficient of repeatability (<5.35).

Relationship with other variables

In our next research question, we asked if there was a relationship between meaningful reading scores and (1) age and (2) standardized measures of receptive vocabulary and reading ability. Our data clearly shows no relationship between meaningful reading score and these other variables. In relation to age, as we stated at the outset, we could have argued for a positive or negative association. Negative, in the context of younger adults with Down syndrome being (1) less likely to experience cognitive decline (McCarron et al., 2017) and (2) less likely to have been educated in a special school and therefore more likely to have increased reading skills (de Graaf et al., 2013). Positive, because of the greater life experiences of older adults which gives them greater exposure to meaningful text reflected in the items in the checklist. It is possible that all these factors were at play (with the effect of one factor negating the effect of another) and therefore no clear relationship emerged. It is also noteworthy that we did not account for cognitive ability in our analysis, a factor that is not independent of our findings.

A lack of relationship between our measure and the standardized vocabulary assessment (PPVT-4) is not surprising and was in keeping with our hypothesis. Most standardized vocabulary checklists are based on a developmental trajectory and tend to reflect vocabulary that might be relevant to primary and post-primary education rather than to socio-cultural experiences in the community. Even tests that have been normed on typical adult populations tend not to include areas relevant to community living, such as popular culture. Vocabulary in typical adults is measured as a construct that increases in difficulty (i.e., levels of abstractness with a focus on more academic language) rather than vocabulary quantity, reflecting a broader range of topics. In contrast, growth in receptive vocabulary development in adolescents and adults with Down syndrome is more likely to be driven by individual, educational, environmental, social, and cultural experiences. Conversations with some of the participants in the current study suggested continuing vocabulary development in areas such as sport, local and national politics and Covid-19, none of which would be reflected in a standardized measure such as the PPVT-4.

It was also unsurprising that there was no relationship between meaningful reading score and The Burt Reading Recognition Test (our standardized reading measure). While some of our participants were unable to read even the first line of the Burt (which consists of the words *to, is, up, he, at*), all participants indicated that they read some of the items on the checklist (a minimum of 12 items). In terms of sight word recognition, words such as t*o, is, up, he, at* tend not to represent the most pertinent information in a sentence and therefore become less relevant to reading key points of information that facilitate functioning in everyday life. The ability to read other words in the Burt (such as *projecting, explorer, domineer*) is reflective of decoding skills without any context and is far removed from reading for a specific purpose where the context provides significant support and the act of reading is underpinned by a different motivation. We did not expect an association between meaningful reading and this standardized word recognition test. As has been previously highlighted in the literature (see Moni and Jobling, 2001), standardized measures like this do not adequately capture reading in a socio-cultural context. Particularly, they do not capture change in how adults perceive themselves as readers. Some participants in the current study described themselves as non-readers at the outset. However, when encouraged to look at the MRM they found the experience to be empowering, and it allowed them to notice the ways that they read the written word in everyday life. Consequently, having completed the measure they began to identify as readers.

Meaningful reading in everyday lives

Our final two research questions addressed what meaningful reading is for adolescents and adults with Down syndrome. Although there was considerable individual variation, all participants indicated that they read some of the items in the MRM (a minimum of 12 and a maximum of 44). Items read, reflected a broad range of reading practices from *restaurant menus, sport fixtures, travel timetables* and *poems* to *lottery numbers* and *wordsearches*. The span of items is indicative of the importance of examining and targeting a range of reading practices within our educational contexts (Street, 2001). In addition, our dataset can guide educationalists designing and developing post-school literacy modules to ensure that programs reflect the full extent of everyday reading practices for this cohort. With respect to where and when individuals with Down syndrome read, our data shows *at home* to be the most popular response and reinforces the idea that reading is an activity that is not confined to educational settings for people with Down syndrome. No clear pattern emerged in relation to when individuals with Down syndrome read, indicating that meaningful reading is integral to people's lives at different times throughout the day. Regarding the most important thing about reading, the majority of responses referred to the fact that reading was an enjoyable activity and that it is something that aids learning. As noted by Williams (2005), pleasure and enjoyment have not been a priority in post-school literacy courses, which tend instead to focus on employment based skills. Despite the fact that it may enhance quality of life, literacy for pleasure and recreation has been neglected (Ashman and Suttie, 1995). Our data reinforces the view that this needs to change. In addition, responses indicating that reading is something our participants did to aid their learning and that they read to access information, demonstrates an ongoing interest in lifelong learning for people with Down syndrome. In keeping with the United Nations' Convention on the Rights of Persons with Disabilities (UNCRPD) it is important that adolescents and adults with Down syndrome are given equal opportunities to access relevant education and training (art. 24) throughout their lives (United Nations, 2006).

Limitations and future steps

There are some limitations to the current study which we note here. Firstly, our information sheets were developed by the research team. Although the team had significant experience working with people with Down syndrome, given the collaborative nature of the work (stage 1) it would have been preferable that our collaborators were integral to this process. Secondly, given that 13 of the 15 images presented to our collaborators (depicting each item) were exchanged, one could argue that we should have asked for feedback on the images that represented all items in the checklist. We did not do this as we believed that some images (such as *receipt*) were easy to depict in a universal manner and we wanted to reduce the burden of the task for our collaborators with Down syndrome. The images we chose for validation were those we deemed to be potentially problematic and we were therefore not surprised that such a large proportion of these were revised. Thirdly, two items were accidentally omitted from the final checklist (i.e., social media and web browsing). Given the number of times they were generated by our collaborators in stage 1, we expect that they would have featured strongly in the items most often read by our stage 2 participants. If it is the case that these items are frequently read by adolescents and adults with Down syndrome, it would support the need to include popular culture in post-school programs, which would serve to build social capital and develop common frames of reference between those with Down syndrome and their "typical" peers (Davies and Dickinson, 2004). Lastly, given that we did not expect a relationship between standardized reading scores and our meaningful reading measure, it would have been preferable to have also included a measure (such as reading engagement) for which a relationship may have been more likely. That said, these measures exist for the general population only and therefore may not be appropriate or may require significant adaptation for people with Down syndrome. As a potential additional validation, we did pilot asking parents to complete the checklist (without picture supports) but it became clear that unless reading books, parents did not view their sons/daughters as readers. It may have been a fruitful exercise to offer "training" to parents in the purpose of the measure and to ask them to observe and document their son or daughter's reading over a specified period.

Regarding future steps, while our sample is representative of a range of adolescents/adults with Down syndrome in Ireland (many of whom attend some form of further education), the sample is relatively small and the measure would now need to be used to profile a much larger group nationally and internationally. To strengthen measurement of change over time it may also be useful to develop a supplemental sheet for family members which could capture frequency data (regarding how often the individual with Down syndrome is engaging with meaningful reading tasks) as well as qualitative information on changes in behavior. This would include the ability to engage in new reading tasks as well as tasks that individuals were previously able to do. Finally, it would be useful to complete longitudinal work to investigate the sensitivity of the MRM in measuring change in reading behavior over longer periods of time.

Conclusion

This study reports on the development of a measure of broad-based reading-related literacy skills in collaboration with a group of adolescents and adults with Down syndrome. Our preliminary data presented here shows that the measure is reliable as well as having strong ecological and content validity. As an outcome measure, the MRM can serve to complement existing standardized tools and can be used to measure change regarding the range of items read pre- and post-reading intervention programs (although further work is required to establish the measure's sensitivity to change over time). The MRM can also guide post-school reading program content to ensure that it is meaningful to the everyday social practices of people with Down syndrome. Lastly, by framing reading as a meaningful daily activity, the MRM can capture growth in these adults' perceptions of themselves as legitimate readers, reflecting an increased confidence and motivation to read. Consequently, it can help educationalists and others in society to recognize adults with Down syndrome as valued literate members of the community in which they live.

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.

Ethics statement

The studies involving human participants were reviewed and approved by the School of Clinical Therapies Social Research Ethics Committee, University College Cork. Written informed consent to participate in this study was provided by the participants' themselves who were given documentation in advance of the study to discuss with their parent/guardian if they wished to.

Author contributions

PF conceptualized the study and wrote the manuscript. SO'D engaged with collaborators to generate the tool and participants to test it (test–retest reliability) and completed some of the standardized vocabulary and reading tests. MJ completed some of the test–retest reliability work and the qualitative data analysis. LM completed some of the standardized tests, facilitated recruitment and assisted with focus group. NH conceptualized the study, facilitated recruitment, completed some test–retest reliability work, assisted with focus group and provided comments on manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2023.1173300/full#supplementary-material

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Narrative abilities in individuals with Down syndrome: single case-profiles

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Introduction: Narrative abilities are an important part of our everyday lives and social interaction with others. Nevertheless, narration is a complex ability influenced by language and cognition. This makes it difficult for individuals with language and cognitive impairment, such as in children and adolescents with Down syndrome. Previous studies have shown distinct narrative impairments in individuals with Down syndrome; nevertheless, this research was based on overall group means in most cases. To identify individual strengths and weaknesses and to draw conclusions for speech and language therapy, the narrative profile of every participant should be considered equally. Following this approach, the current study aims to describe single case narrative profiles in individuals with Down syndrome.

Methods: The narrative transcripts of 28 children and adolescents with Down syndrome (aged 10;0–20;1), based on a non-verbal picture book, were rated using the Narrative Scoring Scheme across seven macro- and microstructural categories. Point scores across the whole group are displayed – nevertheless, the paper specifically addresses the individual narrative profiles of the participants. The participants could be assigned to narrative profile groups which show different characteristics, strengths and weaknesses. Group comparisons and correlations were computed for the relation to language abilities (especially vocabulary) and nonverbal cognitive abilities.

Results: The results of the two profile groups with minimal and developing narrative skills differ significantly not only concerning narrative outcomes in the Narrative Scoring Scheme but also for language abilities and developmental stage of nonverbal cognition. Individuals that show floor effects in narrative abilities are characterized by an overall weakness in language and cognition. In contrast, a group of approximately equal size shows distinct strengths in their narrative profiles which are in line with their vocabulary strengths, MLU and nonverbal cognition.

Discussion: The current study uses a new approach to identify individual narrative profiles in a group of individuals with Down syndrome. The results of the investigation underline the existence of narrative impairments in many individuals with Down syndrome but also point to individual strengths of the participants. Furthermore, the study outcomes suggest that narrative abilities might be representative for overall language and cognition in individuals with Down syndrome. However, intervention studies addressing narration are missing.

KEYWORDS

narration, Down syndrome, single cases, language and cognition, profile analysis

1. Introduction

Narration as a cultural technique plays an important role in social interaction. The social act of narration occurs in the exchange of two or more persons and linguistically comprises the text type narrative. According to Katz-Bernstein and Schröder (2017), this is defined as follows: "[Narratives contain] unique events that contain a special feature, in the form that something unexpected has happened. (...) Quite essential for narration in contrast to reporting is that with the unexpected, the breaking of the plan, an emotional evaluation accompanies it. An important function of narrative is to convey this emotion to the listener" (Katz-Bernstein and Schröder, 2017, p.2, translated by author). Narrative thus goes far beyond functional language and includes formal and communicative parts, as pointed out by Bamberg (2016). Previous investigations have shown a high impact of narrative competences on literacy acquisition (Botting, 2002; O'Neill et al., 2004) and school outcomes (e.g., O'Neill et al., 2004 for mathematical abilities). Regardless of its high relevance in everyday communication, narration is a distinctly complex skill, which is linked to cognitive and linguistic performances, e.g., the Theory of Mind (Tompkins et al., 2019) or the vocabulary of a narrator (Korecky-Kröll et al., 2019; Neitzel, under review). Narratives represent a distinct manifestation of the superordinate expressive form text (Kintsch, 1974; De Beaugrande and Dressler, 1981). In the narrative, successive sentences are linked in terms of content and language – only through this connection does a story emerge. The contextual connection of utterances, the so-called coherence, enables listeners to connect to their own prior and world knowledge (De Beaugrande and Dressler, 1981) and serves as content orientation within a narrative (Swoboda, 2011). This usually follows a conventionalized, recurring form, the so-called macrostructure. This macrostructure is a formal blueprint of a story, which according to Stein and Glenn's (1979) story grammar model consists of six grammar units (setting, initiating event, internal response, attempt, outcome, response; detailed introduction in Govindarajan and Paradis, 2022). Within a story, there can be so-called story episodes, i.e., different plot lines or events, which are, however, each structured according to the story grammar model. The three grammar units initiating event, attempt and outcome are obligatory, while other elements can be variably located in their position (internal reaction of characters) or optionally added (morality; Van Dijk, 1980). Macrostructure is thus a formal structure that is realized across content. Impairments of the macrostructure, which can occur in the context of developmental language disorders, manifest themselves, for example, through missing grammar units or an unstructured narrative sequence. Microstructure, on the other hand, "refers to a local level of analysis" (Govindarajan & Paradis, 2022, p. 1363) and includes all concrete linguistic information, e.g., word choice or sentence length. The concrete linguistic realization of narrative content is implemented at the microstructural level through the use of cohesive devices. These include, for example, conjunctions – which connect individual sentences – or pronouns, which enable references across sentences. Overall, microstructure encompasses "measures of word frequency, proportion of content words (i.e., nouns and verbs), grammaticality and sentence complexity" (Altman et al., 2022, p. 1119). Impaired language abilities can manifest themselves on a microstructural level, for example, through morphological or syntactic errors or an insufficiently differentiated vocabulary.

The complex interplay of different language levels and cognitive abilities, as well as the demands of the narrative form itself, lead to limitations in narrative ability. These have been observed in different populations. Narrative abilities of speakers with Down syndrome – a group of individuals characterized by multiple cognitive and language disabilities – have already been investigated in the literature by various research groups. Due to the extent of previous findings and research designs, a complete literature review is not possible here; reference is made to, among others, an extensive review by Segal and Pesco (2015). However, the current state of research can be clustered based on the following assumptions and the respective methodological focus: A large part of the international research concludes that speakers with Down syndrome show strengths in the macrostructure of a narrative (Keller-Bell and Abbeduto, 2007; Finestack et al., 2012; Segal and Pesco, 2015). In parallel (and rarely overlapping methodologically), some studies focused on the (underlying) language impairments of narrators with Down syndrome, highlighting impairments primarily at the microstructural level (Finestack et al., 2012; Channell et al., 2015; Ashby et al., 2017). This is illustrated subsequently on the basis of selected studies.

A study by Finestack et al. (2012) illustrates a comprehensive assessment of the narrative performance of speakers with Down syndrome. It focused on both macro- and micro-structural levels, and included a comparison group of typically developing children aged 4–6 years. They surveyed the narrative ability of 24 English-speaking adolescents and young adults (chronological age: $M = 16;11$ years, SD 3;2 years, range 12;1–23;4 years; mental age: $M = 4;11$ years, $SD = 1;0$ years, range 3;4–7;1 years) using the Narrative Scoring Scheme (NSS, Heilmann et al., 2010; see section 2.2). In an individual matching of (non-verbal) mental age between participants with Down syndrome and the control group ($n = 21$), the speakers with Down syndrome showed a macrostructure appropriate for their non-verbal mental age and significantly outperformed the typically-developing participants in terms of the macrostructure element introduction as well as the total score. Similarly, Neitzel and Penke (2022a) were able to show in a profile comparison of children with typical development and participants with Down syndrome with a mean non-verbal mental age of 5;03 years (in y;mm) that the narrative performance of the participants with Down syndrome – measured by NSS-scores – corresponded to that of 5-year-old typically developing children on group average. Such findings contribute to the widespread assumption that narrators with Down syndrome might show relative confidence in macrostructure (Segal and Pesco, 2015) – as measured by non-verbal cognitive stage of development, not chronological age.

In contrast, the results on microstructural performance in people with Down syndrome are more equivocal, which may be partly due to the methodological approach. Many studies in the literature have used MLU as a microstructural measure of narrative ability. Nevertheless, there are mixed findings in the literature on MLU in narratives of participants with Down syndrome. MLU is repeatedly used as an overall measure of grammar in narrative studies (e.g., Ashby et al., 2017; Channell, 2020), with high MLU indicating higher grammar skills. Neitzel and Penke (2021), in contrast, were able to show that higher MLU in participants with Down syndrome may rather be a manifestation of syntactic impairment. In their study, the MLU of participants with Down syndrome was even slightly higher than the MLU of a 9-year-old comparison group of typically developing children, despite a mean mental age of 5;03 years. This finding, however, was not caused by a high syntactic complexity, but was an expression of long but syntactically incoherent sentences. Accordingly, MLU is an important covariate concerning the morpho-syntactic abilities of individuals with Down syndrome in research, but great caution must be exercised in interpreting the pure values. The assumption that a higher value automatically indicates higher grammatical abilities does not apply unreservedly to these participants. The extent to which looking only at the group mean of the MLU can be misleading in the interpretation of narrative performance, can be demonstrated by the study of Finestack et al. (2012), in which participants were compared with typically-developing children of the same non-verbal mental age. The group mean in MLU of the two groups was comparable (differences n.s.). Strengths of the participants with Down syndrome were evident in the macrostructure, with these participants even outperforming the typically-developing children in some cases. However, when participants from both groups were matched 1:1 according to their MLU, no differences in favor of the participants with Down syndrome were detectable anymore. Firstly, this indicates that the group mean in MLU led to a distorted picture of individual performance. Secondly, this points to a strong interaction between macro- and microstructure, which makes it methodically difficult to differentiate between both constructs. These critical points lead to concerns regarding the interpretation of narrative performance on group level in individuals with Down syndrome. Overall, the presented research overview provides a partly ambiguous picture of narrative abilities in persons with Down syndrome.

Group evaluations, especially the focus on group mean comparisons and significances, have their value for basic deduction in narrative research. At the same time, the existing research lacks a focus on the individual performance of the participants, although individual aims, e.g., for speech and language therapy, can only be chosen on a case-by-case basis. Inferences from group results to the individual case may leave individual strengths and weaknesses undiscovered. Particularly speaking about narrative performance, which encompasses a wide range of abilities, group means are not necessarily informative about what individual narrative support for children and adolescents with Down syndrome should look like. Therefore, the present focus on single cases is primarily intended to provide clinical and educational conclusions and to demonstrate an exemplary approach to making research on individuals with Down syndrome more individualized. The investigation presents an analysis of individual performance profiles with a focus on the question of whether definable subgroups and competence profiles emerge in this context. The assessment should produce an overarching narrative profile for each case, encompassing macro- and micro-structural aspects of narrative competence, both of which are essential to narrative. For this reason, an assessment tool (Narrative Scoring Scheme, *cf.* 2.2) is used which allows for an overall view per case (total score), but at the same time allows for the derivation of individual support approaches on a case-by-case basis (identification of resources and weaknesses). At the same time, however, group performances are presented, in order to classify the narrative abilities of the examined children and adolescents with Down syndrome against the background of past research results.

2. Methods

2.1. Participants

Twenty eight children and adolescents with Down syndrome participated in the current study (free trisomy: $n = 26$; mosaic trisomy: $n = 1$; type of trisomy unknown: $n = 1$). All participants were recruited from institutions such as special needs schools and inclusive sports clubs[1] with the aid of parent associations. The participants with Down syndrome were monolingual German speakers (15 f., 13 m.) and attended an inclusive ($n = 17$) or special needs school ($n = 11$). All individuals were Caucasian. The educational level of the families was variable, but high overall (15 mothers and fathers each with university entrance qualifications and/or academic degrees). Sufficient ability in hearing and vision was reported for all individuals (unimpaired hearing in $n = 19$ participants, mild hearing loss 10–30 dB in $n = 9$ participants). Participant characteristics and outcomes from cognition and vocabulary measures (see section 2.3) as well as MLU (see section 2.2) are presented in Table 1. The sample consists of older children, adolescents, and few young adults with Down syndrome (chronological age: $M = 14;05$ years;months). In the cognition test SON-R the participants scored on average 59.93% of the points and in the expressive vocabulary test AWST-R (description of both tests in 2.3) 66.28% of the points. The MLU of the participants is high ($M = 7.00$), but this is due to syntactic impairments in many participants (Neitzel and Penke, 2021). The research project involving the data presented here was approved by the Ethics Committee of the Medical Department of the University of Cologne (number of approval 18–121). Inclusion criteria for participation were growing up monolingual and verbal utterance skills at least at two-word level.[2] Section 2.4 describes the procedure administered in the study.

2.2. Narrative measures

The present evaluation of narrative abilities was conducted on the basis of written transcriptions of the narratives that the participants produced using the so-called Frog Story. The Frog Story is a nonverbal picture book ('Frog, where are you?'; Mayer, 2003), including 24 black and white illustrations, that is widely used in research on narrative abilities. The required noun vocabulary for the story (acting characters and central objects/locations) was secured in advance using picture naming in a prepared PowerPoint presentation.[3] The Frog Story as well as the scoring procedure (see next paragraph) has already been used successfully with participants with Down syndrome by Finestack et al. (2012). The research procedure, which is described below, is internationally common in this form and goes back to a study by Reilly et al. (2004). This approach could be used congruently for the participants with Down syndrome. The Frog Story picture book was

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[1] Inclusive sport clubs are a leisure activity that can be attended independently of schooling in Germany. In this case, it is an association where parents can have their children with Down syndrome cared for on weekends as part of preventive care.

[2] Mental age, as calculated using SON R 2 ½-7 (see section 2.3)>3;6years was another inclusion criterion. Raw scores from the SON-R are presented in Table 1, whereas mental age is not part of the current analyses.

[3] The occurring nouns were presented to the participants as separate picture details and were to be named by the children. If a child was unsure about an item or named it incorrectly, the experimenter named the word correctly and spoke briefly with the child about the respective item. At the end of the review, this noun was then asked again. The children were able to name the core vocabulary of the respective stories in all cases at least in the second run. Afterwards, the children began to look at the original book.

TABLE 1 Participant characteristics concerning age, cognition, vocabulary and MLU across the group.

Instrument	Chronological age (in y;mm)	SON-R 2 ½-7 (Cognition) Reasoning Scale Raw score (max. 46 p.)	AWST-R (Vocabulary) Raw score (max. 75 p.)	Frog Story MLU (in words)
Mean	14;05	27.57	49.71	7.00
SD	2;06	6.18	13.44	2.94
Range	10;00–20;01	17–42	12–64	1.57–13.28

introduced by the experimenter ('Look, I have this book for you. I want you to tell me a story about it in a moment. First, let us look at the book together. Just look at the pictures.'). The book was presented nonverbally by the experimenter, who slowly leafed through it in a way that was clearly visible to the child. For each illustration, the child was given sufficient time to look at it, but no linguistic request was made yet. Subsequently, the participant was asked to tell the story on the basis of the illustrations. To do this, the book was flipped through page by page again, with the experimenter using only non-specific questions such as 'What is happening here?'

Written transcripts of the narratives were made using ELAN 5.3 (Max Planck Institute for Psycholinguistics, The Language Archive, 2018). Transcription was controlled by two additional, individual raters (trained student assistants). Disagreements were discussed and resolved through a consensus process. Intelligibility of the individuals was partly limited by phonologic errors but did not affect the narrative analyses.[4] The participants' narrative ability was evaluated using the Narrative Scoring Scheme (NSS; Heilmann et al., 2010) in seven subcategories with zero to five points (max. 35 points). The NSS allows for the assessment of narrative performance using predetermined categories and a point scoring system and is widely used in narrative research. For the Frog story, comprehensive scoring examples are available to identify an immature (1 point), developing (3 points), and mature (5 points) narrative performance per subcategory (Miller et al., 2003).[5] This handout by Miller et al. (2003) has since been translated into German in an expanded form and is freely available (Neitzel and Meier, 2023). For each category, the handout indicates exactly for which narrative content and linguistic features which score is to be assigned. Neitzel and Meier's (2023) manual was evaluated in a study on transcripts from 89 typically developing children. For example, this may look like the following for the "conclusion" category: For this category, three central 'events' have been named which characterize the content of the story's ending (Miller et al., 2003) – (a) The boy and the dog find the frog, (b) The boy takes a baby frog as a pet, (c) The boy waves/says goodbye and is happy. The manual indicates exactly how many points may be awarded each time a certain number of events are mentioned, e.g., 0 points for no event, 1 point for one event, 3 points for 2 events, 4 points for 3 events. In addition, 2 points are awarded if 1–2 events are mentioned and the end of the story is abrupt but clear (e.g., by the phrase "And over."). Five points are awarded if the narrative is completely rounded off, possibly by common (German) phrases such as "And if they did not die, they are still alive today." Thus, the evaluation can be done very specifically by trained raters. An interrater review revealed a very good reliability of 0.93 (95% CI: 0.77–0.98; Meier and Neitzel, 2023). Note that Heilmann et al. (2010) originally described the NSS to be a macrostructural instrument (clear for subcategories introduction, mental/emotional states, conflict/resolution, cohesion, and conclusion assess macrostructural skills). Nevertheless, the categories of character development – where choice of words is really important – and referencing – which interferes with grammar abilities by scoring, e.g., sentence linking – are more associated with the microstructure of a story. In the current investigation, core microstructural measures are number of different verbs and MLU. Nevertheless, NSS-scores should be considered as a combination score evaluating macro- and microstructural abilities.

2.3. Further standardized measures

The children's and adolescents' cognitive abilities were assessed using the reasoning scale of the SON-R 2 ½-7 nonverbal intelligence test (*Snijders-Oomen Non-verbal intelligence test-revised*; Tellegen et al., 2007). This instrument includes three subtests with 46 items in total: categories, analogies, and situations, and allows participants to respond both verbally or nonverbally. The reported reliability for the reasoning scale is 0.83 (Tellegen et al., 2007). The test is normed for a developmental age of 2;6 to 7;11 years (years; months). The SON-R has been used successfully in many studies with participants with Down syndrome (including Witecy and Penke, 2016). Since the instruction and response of the children can be non-verbal, no adaptation of the implementation was necessary. With respect to their language comprehension abilities, all participants were able to understand the instructions for the measures used.[6] The vocabulary abilities of participants with Down syndrome were assessed by applying the AWST-R (*Aktiver Wortschatztest für 3- bis 5-jährige Kinder*;

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[4] Full narrative transcripts of all participants with Down syndrome are available at CHILDES database: https://childes.talkbank.org/access/Frogs/German-Neitzel.html.

[5] The intermediate values 2 and 4 points were defined within the project and applied comparably by all raters. The interrater reliability for the evaluation of the transcripts, which was carried out on the basis of this manual, could be estimated as very high in a study by Meier and Neitzel (2023) [intraclass correlation on the basis of the two-factorial unadjusted random model, 0.93 (95%-CI: 0.77–0.98)].

[6] Instruction on the measures used was given using simple SVO sentences (word order in German: subject, verb, object). Instructional comprehension was secured as part of the overarching research project by use of a standardized grammar comprehension measure (German version of the TROG). All children passed the first comprehension blocks in which simple sentence structures are tested.

Kiese-Himmel, 2005), a widely used German productive vocabulary test containing 75 items (51 nouns, 24 verbs), presented with increasing difficulty and normed for three- to five-year-old typically developing children. The internal consistency of the AWST-R is α = 0.88 (Kiese-Himmel, 2005). A possible adaptive approach to test administration and scoring for participants with Down syndrome is described in Neitzel et al. (2021). For the present sample, however, no adaptation was necessary compared to the manual-faithful implementation.

2.4. Procedure

The participants were tested as part of a research project on the narrative skills of people with Down syndrome at University of Cologne. The participants took part in three test sessions of 45 to 60 min each. Written parental consent was obtained beforehand. In addition, at the first appointment, parents and child were verbally informed and a parent questionnaires on developmental history was handed out. At the first appointment, in addition to contact games, the SON-R 2 ½–7 (non-verbal cognition) and the expressive vocabulary test AWST-R were administered. In the second session further tests, mainly morpho-syntactic, were administered which are not part of this paper. The frog story narratives were collected in test session three. Each of the sessions was interrupted by appropriate rest breaks. Most of the testing took place in the participant's home environment and some in the institutional environment (school). However, for each participant the testing location was kept constant across all three sessions. Participants were given a small, age-appropriate gift (e.g., sweets and pens) as a thank you for their participation in the study. Parents were also given a detailed report of their child's test performance to give to their child's speech and language therapist or teacher.

2.5. Data analysis

Results for the standardized measures were calculated according to the manual. Only raw scores were used in the present analyses. MLU in words as overall grammar measure and number of different verbs as a measure of verb vocabulary were calculated on the basis of the written transcripts of the narratives. Data processing in the current study was conducted using *SPSS 28* (IBM Corp, 2021). Analyses included the following steps: First, each participant's narrative profile was manually assessed by two independent raters using the NSS. Mean, *SD* and range were calculated for each NSS category and total score. According to this profile (individual scores in the NSS categories), the participants were manually assigned to profile groups, which differed according to the distinguished ability levels of the NSS: minimal ability (1 p. in mean), developing ability (3 p. in mean) and advanced ability (5 p. in mean). Pearson's correlations were used to examine the relationship between NSS scores (total score) and performance on various language and cognitive measures. In addition, non-parametric group comparisons were computed with respect to the language and cognitive variables and narrative performance (Mann–Whitney *U*-tests). To allow for a better understanding of the case profiles, individual associations and dissociations between NSS scores and language or cognition measures were considered using median split analyses and exact Fisher tests in each participant. The total score for the sample is reported in the results section and compared to group studies from the literature in the discussion to allow for classification despite the single case focus.

3. Results

3.1. Narrative group overview and profile groups at single case-level

Following the objective of the present study to not only rely on group means when assessing the narrative abilities of children and adolescents with Down syndrome, but to focus more on individual performance, each individual with Down syndrome was assigned to a profile group according to her narrative abilities. In addition, the Supplementary material of this article provides a complete overview of the narrative performance (NSS subpoints per category) provided by each individual case. The categorization that is typically made in the NSS was selected as a more reliable criterion, namely the differentiation between an immature (1 point), developing (3 points), and mature (5 points) narrative performance. A profile group was therefore created for a participant's mean score of 1, 3, and 5 points in narrative performance. This resulted in the following distribution: Profile group (1) including participants showing minimal narrative abilities with a point score per subcategory of 0 to 1.99 points ($M = 1$); group (2) including participants showing developing narrative abilities defined by a category score of 2.00 to 3.99 points ($M = 3$); group (3) including participants showing advanced narrative abilities with a category score of ≥ 4 points ($M \geq 4$). Since a mean value of 5 points can hardly be achieved in purely mathematical terms due to a maximum 5 points per category, $M \geq 4$ was set as the criterion for profile group (3). Sixteen individuals could be assigned to profile group (1) – minimal narration, MN – and 12 individuals met the criteria of profile group (2) – developing narration, DN – while no participant could be assigned to profile group (3). Table 2 shows the mean NSS-scores of the participants in each profile group in addition to the total group. The results consistently show a higher point score in the DN group than in the MN group in all individual categories and in the total score. According to the mean values, the introduction ($M = 3.83$) and the cohesion ($M = 3.17$) are strengths of the DN group. However, in line with the results of the MN group ($M = 0.75$), the participants in the DN group also achieved low scores ($M = 1.25$) in the subcategory 'conflict/resolution'. As no participant in the sample reached more than 2 points, this category was not included in the assignment to the profile groups. Regarding the present forms of trisomy ($n = 26$ participants with free trisomy, $n = 1$ each with unknown and mosaic trisomy), it can be reported that the participant with unknown form of trisomy was located in the profile group MN. The participant with mosaic trisomy showed a performance corresponding to the DN profile group.

The number of participants in the respective profile group indicates that a comparable number of individuals show a minimal and developing narrative performance whereas no participant can be categorized as a strong narrator. Figure 1 contrasts the individual narrative profiles of all participants from the two groups (please see full narrative profiles per case in Supplementary Table S1). It is clear that in group DN, there are significantly more downward deflections (individual weaknesses) than upward deflections (strengths). A few participants from group MN also show individual scores that exceed three points, but this is always only a single category per participant that exceeds the two-point mark.

TABLE 2 Mean NSS-scores (SD; range) for profile groups of individuals with Down syndrome concerning narrative abilities with 0–5 points (p.) per subcategory.

Mean (*SD*; Range)	Total group (*n* = 28)	Profile group (1): minimal narration (MN) (*n* = 16)	Profile group (2): developing narration (DN) (*n* = 12)	Profile group (3): advanced narration (*n* = 0)
Introduction (0–5 p.)	2.71 (1.41; 0–5)	1.88 (1.20; 0–4)	3.83 (0.72; 3–5)	--
Character Development (0–5 p.)	1.79 (1.13; 0–5)	1.19 (0.66; 0–2)	2.58 (1.16; 1–5)	--
Mental/emotional states (0–5 p.)	1.61 (0.92; 0–4)	1.13 (0.50; 0–2)	2.25 (0.96; 1–4)	--
Referencing (0–5 p.)	1.96 (1.14; 0–4)	1.25 (0.86; 0–3)	2.92 (0.67; 2–4)	--
Conflict/ solution (0–5 p.)	0.96 (0.58; 0–2)	0.75 (0.58; 0–2)	1.25 (0.45; 1–2)	--
Cohesion (0–5 p.)	2.25 (1.08; 0–4)	1.26 (0.81; 0–3)	3.17 (0.55; 2–3)	--
Conclusion (0–5 p.)	1.79 (0.96; 0–4)	1.38 (0.62; 0–2)	2.33 (1.07; 1–4)	--
NSS total score (max. 35 p.)	13.07 (5.90; 1–26)	9.13 (3.67; 1–13)	18.33 (3.77; 14–26)	--

No participant in the sample reached more than 2 points in the subcategory 'conflict/resolution.' Therefore, this category was left out of the assignment to the profile groups.

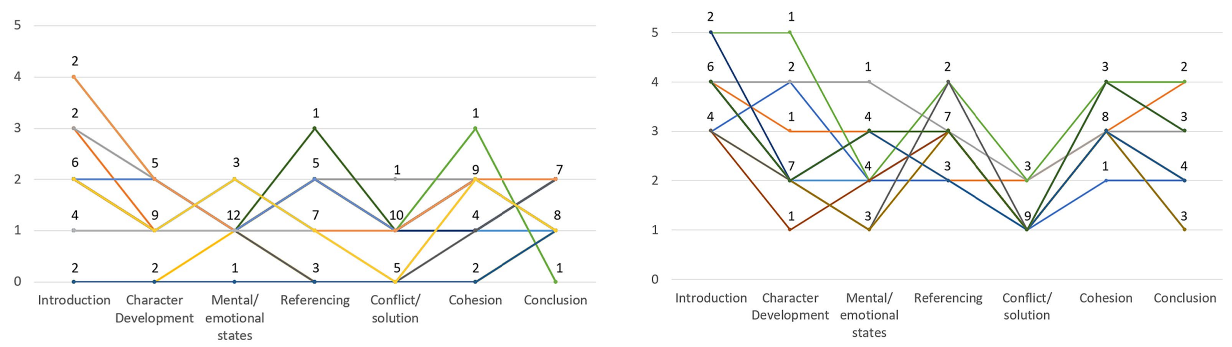

FIGURE 1
Overview of participants in narrative groups minimal narration (MN, left) and developing narration (DN, right). Each line represents one individual participant. The data labels indicate in each case how many participants have reached this value. For participants with an overlapping curve section, only a single line is displayed for visual reasons. Full narrative profiles per case are displayed in Supplementary Table S1.

3.2. Relations to cognition and language abilities

The presented narrative profiles show differing competencies concerning narrative abilities in children and adolescents with Down syndrome. The question remained open whether the individual assignment to these profile groups might be reflected by cognition or language abilities of the individuals. The relation between these factors was investigated in the subsequent analyses.

In this context, correlation analyses were first used to investigate which characteristics and abilities of the participants are associated with narrative abilities. Included here were the children and adolescents' chronological age, cognition outcomes (SON-R raw scores) and MLU as overall grammar measure, as these have been consistently associated with narrative abilities in individuals with Down syndrome in the research literature (e.g., Finestack et al., 2012; Hogan-Brown et al., 2013; Channell et al., 2015). Based on recent findings that narrative abilities in individuals with Down syndrome are highly associated with vocabulary performance (Neitzel and Penke, 2022b) and specifically verb vocabulary (number of different verbs; see analyses in Neitzel, under review), these two expressive vocabulary measures were included. Table 3 shows the results of these analyses, which display significant correlations ($p < 0.05$/$p < 0.001$) between NSS-total scores and all measures except from chronological age.

Since the correlation results did not provide any insight whether individuals in the two profile groups – profile group (3) (advanced performance) was disregarded at this point, as no participant met the criteria for it – differed in terms of their performance on the variables included (see Table 4), the performance was analyzed per profile group. The results indicate that the narrative ability profiles indeed reflect performance in other language and cognition measures. Accordingly, a non-parametric group comparison (Mann Whitney U) revealed a significant difference for cognition raw scores, vocabulary measures and MLU between the two profile groups (each $p < 0.001$). The number of different verbs used in the narratives was also significantly higher among participants in the DN group ($M = 34.83$) than in the MN group ($M = 23.31$; $p = 0.006$). At the same time, the examined participants of the two profile groups do not differ with regard to their chronological age and thus regarding their language experience.

To investigate the narrative performance of participants in the two profile groups more deeply regarding their cognitive and language abilities, a median-split analysis was conducted to compare associations and dissociations between sub-median performance and above median performance on each measure and the assignment to group MN or

TABLE 3 Correlations of narrative abilities (NSS-score) and age, cognition or language variables for all participants ($n = 28$).

	Total NSS-score	Chronological age	Raw score cognition	Raw score expr. vocabulary	*n* different verbs in Frog story	MLU in words
Total NSS-score		0.262	0.706**	0.841**	0.736**	0.721**
Chronological age			0.658**	0.299	0.137	0.154
Raw score cognition				0.681**	0.437*	0.584**
Raw score expr. vocabulary					0.627**	0.614**
n different verbs in Frog story						0.578**
MLU in words						

Difference is significant for *$p < 0.05$, **$p < 0.001$.

TABLE 4 Mean results (SD, range) concerning age, cognition and language variables across profile groups and results of non-parametric group comparison (Mann–Whitney *U*).

Mean (*SD*; Range)	Total NSS-score (max. 35 p.)	Chronological age (in y;mm)	Raw score cognition (SON R 2 ½-7) (max. 46 p.)	Raw score expr. Vocabulary (AWST-R) (max. 75 p.)	*n* different verbs in Frog story	MLU in words
Profile group minimal narration (MN, $n = 16$)	9.13 (3.67; 1–13)	13;7 (2;4; 10;00–18;8)	24.06 (4.43; 17–31)	49.71 (13–67; 12–58)	23.31 (10.23; 4–37)	5.49 (2.67; 1.57–10.83)
Profile group developing narration (DN, $n = 12$)	18.33 (3.77; 14–26)	15;6 (2;7; 11;0–20;01)	32.25 (5.01; 25–42)	58.83 (5.46; 45–64)	34.83 (7.28; 25–48)	9.02 (1.83; 6.7–13.2)
Group comparison (*p*-values)	<0.001*	0.066	<0.001*	<0.001*	0.006*	0.001*

*Difference is significant for $p < 0.05$.

DN. The data for this comparison is visualized in Figure 2. Concerning their performance in the standardized expressive vocabulary measure AWST-R, 16 participants reached scores < median (< 54,00 points) and 12 participants showed scores ≥ median. Remarkably, the number is equal for the cognition measure (SON-R, median score 27.5) and MLU (median score 7.14). Given that 16 participants were assigned to group MN following their NSS-score and 12 participants to group DN, their narrative performance seems to be reflected very closely by sub-median or above median performance in the cognition and language measures. Concerning associations and dissociations, a comparable image is displayed for every measure: whereas 22 participants display associations between a lower or higher performance in the cognition/language measure and their narrative ability (e.g., vocabulary < median & group MN or vocabulary ≥ median & group DN), 6 participants display a dissociation between these measures. 5 participants were assigned to group DN although they show vocabulary skills < median, while one participant was assigned to group MN and showed vocabulary skills ≥ median. In the cognition measure and MLU, 4 participants display sub-median performance and were still assigned to group DN, whereas two participants show above median performance but low narrative skills (group MN). Exact Fisher tests for all comparisons are significant (vocabulary: $p = 0.002$, cognition & MLU each: $p = 0.006$).

4. Discussion

The present study investigated narrative skills based on stories collected from 28 children and adolescents with Down syndrome using a non-verbal picture book. In comparison to previous studies, this study did not focus on group mean analyses, but assessed each child's individual narrative profile. The point scores, that are differentiated in the NSS instrument, have been used as an orientation for the formation of narrative ability profile groups, which ranged from minimal to developing abilities and showed similarities with the children's and adolescents' performance in other language and cognitive domains. The detailed results of the present study are discussed below.

4.1. Narrative performance in individuals with Down syndrome

To allow for a general understanding of the data of the current investigation, the narrative abilities of individuals with Down syndrome are briefly discussed on group level (overview in Table 2). Results are classified with regard to previous studies, especially the NSS-scores described by Finestack et al. (2012). Afterwards, the data

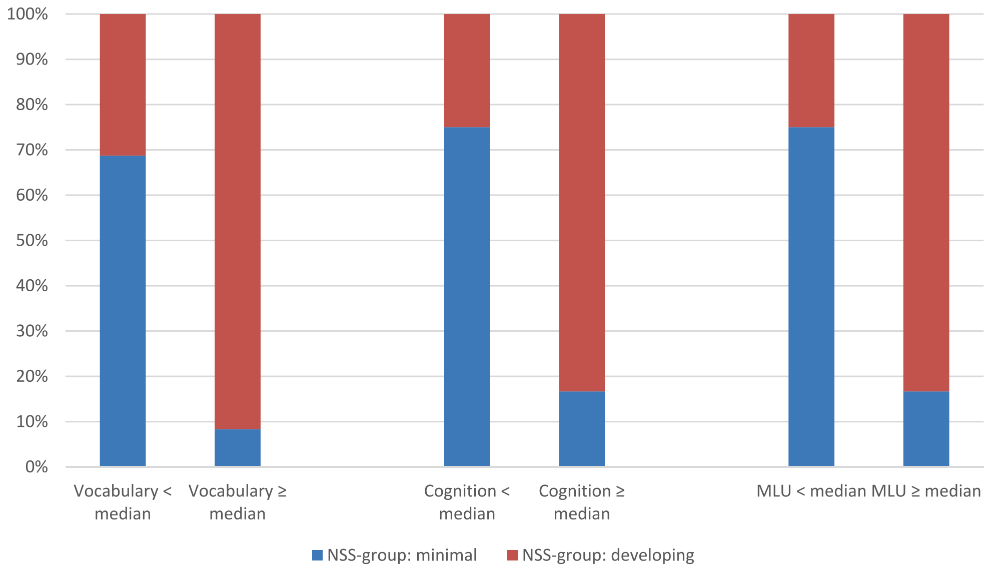

FIGURE 2
Individual associations and dissociations between language/cognition measures and narrative performance (NSS-score groups minimal narration (MN) and developing narration (DN)) (median splits).

are discussed on a single-case level (with regard to the defined profile groups).

4.1.1. Group level

The group results indicate that children and adolescents with Down syndrome show a significant limitation in the area of storytelling skills. Although the Frog Story could be performed with all participants, there was a wide point range in the categories of the NSS. This underlines a considerable heterogeneity concerning performance in the narrative task. While some participants were able to convey very little narrative content and – in some cases – scored zero in several categories, there were also 42.9% of individuals ($n = 12$) who displayed a developing narrative performance with an average of three points in the NSS. However, the majority of participants ($n = 16$, 57.1%) scored less than two points per category on average. In accordance with the scoring guidelines of the NSS, this points to an immature or minimal narrative ability in the early stages of development. Neitzel and Penke (2022a) were able to show that, despite these limitations, the performance of the current sample (in mean) corresponded to the performance of 5-year-old typically developing children and was, thus, appropriate for their calculated mental age of 5;03 years on average. At the same time, the results showed a higher range than those of typically developing children, which is one reason why this paper aims at a more in-depth, case-by-case analysis of narrative skills (see 4.1.2). Methodically, it is important to underline in this context that the comparison of individuals with Down syndrome and typically-developing children must be handled very carefully and is often criticized, which also includes the use of (non-verbal) mental age for matching-procedures. In the current analyses, no mental age was therefore used as a benchmark for narrative or other developmental levels. Nevertheless, previous analyses such as the findings from Neitzel and Penke (2022a) involved this method and are therefore reported accordingly.

While the categories introduction and cohesion represented a relative strength of the participants, the children and adolescents with Down syndrome only achieved an extremely low mean score of 0.96 points (range 0–2 points) in the category conflict/resolution. This suggests that individuals with Down syndrome are often unable to realize the central conflicts and resolutions of a narrative linguistically, which can significantly impede listeners' understanding of the story. While the finding that subjects with Down syndrome scored relatively high in the introduction and cohesion categories is consistent with the findings of Finestack et al. (2012), the participants in the current study showed a distinctly lower score in the conflict/resolution category than was the case with Finestack et al. (2012) (group mean 13.07 vs. 17.21). This may suggest that the distinct impairment in the important narrative feature of realizing conflict and resolution, that the participants showed here, cannot be generalized for all individuals with Down syndrome. It is also possible that the differences could have arisen during implementation, for example, if the Finestack et al. (2012) study had provided assistances that were not detailed in the paper. Alternatively, the differences could have occurred during the scoring process, as the NSS recommendations by Heilmann et al. (2010) only provide guideline scores for items 1, 3, and 5, which are orientational in nature. The point values 2 and 4 could therefore have been assigned according to different criteria, likewise the point value 0. The discussed scores on the (macrostructural) NSS subcatgories introduction, cohesion, and 'conflict/resolution' indicate both strengths and weaknesses regarding the macrostructure of a story in the studied participants with Down syndrome. At first glance, this seems to contradict the assumption of other authors that individuals with Down syndrome often show strengths at the macrostructural level (e.g., Keller-Bell and Abbeduto, 2007; Finestack et al., 2012; Segal and Pesco, 2015). However, since the category 'conflict/resolution' might be a difficulty for typically developed children as well, as examined by Neitzel and Penke (2022a), it also seems possible here that this category represents a particular complexity for storytellers in the learning process. Therefore, this category may need to be further explored in future studies in order to distinguish possible syndrome-specific difficulties of people with Down syndrome from an (expected) delay in narrative skills. The fact that this study cannot specifically address macro- and microstructure due to its focus on an overarching narrative profile of people with Down syndrome is, however, a limitation of this paper.

The remaining ambiguities as well as the great standard deviations in the category results indicate that group comparisons are only of limited use to represent the narrative performance of individuals with Down syndrome. Moreover, they do not allow for individual derivations of clinical implications. The following discussion of individual cases is intended to make this possible.

4.1.2. Single case-level

As described, each participant exhibits a different and individual narrative profile in the current investigation. At the same time, certain profile groups seem to emerge, which allow a rough orientation concerning the narrative performance level; these profile groups are remarkably reflected in the linguistic abilities and the non-verbal developmental level, measured by SON-R raw scores. A large proportion of participants ($n = 12$, MN group) showed minimal narrative abilities, which were accompanied by limitations in vocabulary (weaker performance as measured by the other group) and lower MLU. Since MLU is often distorted in participants with Down syndrome, in this case it cannot be assumed *per se* that the individuals produced shorter utterances than the other participants with Down syndrome (see detailed discussion of the relation to linguistic and cognitive factors in section 4.2). However, there is a significant difference from the group with developing narrative abilities (DN) in terms of performance in the nonverbal cognition measure. A lower developmental level in nonverbal cognition would be a possible explanation for the occurring floor effects in group MN. However, because of the overall relatively low language performance (as measured by vocabulary measures and MLU), an alternative hypothesis would be that these participants show the most severe limitations in their linguistic-cognitive profile among the participants of the study, and that the low narrative abilities might be only one of several impairments. It is a limitation of the present study that no comparison data from other populations with intellectual disabilities was obtained that would allow a more precise interpretation of the results.

The present results provide a first insight that the striking heterogeneity in the narrative performances of participants with Down syndrome – as measured by group means in previous studies, e.g., by Finestack et al. (2012) – could possibly be explained by different narrative profiles and developmental stages. In this context, it would also be helpful to conduct intervention studies that could help to examine the skills of individual groups. On the other hand, these could also be used to implement an even stronger focus on the individual case, since the profile groups presented here naturally also represent a form of clustering.

The analyses presented here shed light on narrative performance and possible profiles of individuals with Down syndrome and thus not only open up further research areas, but also point to clinical implications. Even if the presented results are only a rough orientation and explorative in nature (see also limitations in section 4.3), the different narrative profiles indicate different developmental levels in storytelling skills. Since narration is an important basic ability of interaction in our everyday life, work on the narrative level should not be left out in participants with Down syndrome. It is necessary to examine in the form of further investigations – above all in intervention studies – which concrete starting points result from the individual performances.

4.2. Associations of narrative abilities and language or cognition variables

In the last step, this study investigated whether the cognitive and language impairments of individuals with Down syndrome are related to their narrative abilities. Previous research suggests that narrative abilities in individuals with Down syndrome might be related to cognition, vocabulary performance and MLU. These measures were therefore considered as possible factors influencing narrative performance in speakers with Down syndrome in this context. The correlation analyses performed show a clear correlation between narrative performance, measured by the NSS score, cognition and all language measures. An exception is chronological age, which shows no significant correlation. In this regard, the results underline that chronological age might be no determining factor for language performance in individuals with Down syndrome (evidence of exceptions exists, e.g., for grammar comprehension, see Witecy et al., 2021). Furthermore, the results point to the many factors and skills involved in a (successful) narrative. The individual linguistic and cognitive skills are nevertheless interrelated according to the correlation analyses, which makes a clear picture of directional connections difficult. Moreover, with regard to the individual case-oriented evaluation, the correlation analyses do not provide any insight.

The individual cognitive and linguistic measures were therefore considered separately for the different defined profile groups. Non-parametric group comparisons reveal significant differences in all cognitive and language measures except chronological age. The two profile groups are thus comparable in their mean chronological age, but independently show completely different language and cognition profiles. For all measures considered, a clear difference in favor of individuals in the DN group concerning the achieved values can be observed, which indicates a better linguistic performance and a higher stage of cognitive development in the respective participants. The difference is very pronounced for MLU, but due to the morpho-syntactic impairments in many individuals with Down syndrome, the interpretation of this difference should be made with caution; thus, since the MLU of the participants is high overall across the individual profile groups, this does not necessarily indicate lower performance in the MN group. Rather, it must be remembered that a higher MLU of the participants in many cases occurs due to syntactic deficits, for example, sentence entanglements. Neitzel and Penke (2021) were able to show in a syntactic analysis for the sample presented here that the participants showed a high degree of sentence fragmentation. These are evidence of the syntactic deficits present in many participants. Likewise, the analyses showed the described sentence entanglements, for example in the following sentence: "*Wir wissen noch nicht was sind die beiden was meint.*" 'We do not know yet what the two are meaning' (Neitzel & Penke, 2021, p. 8).

A median split analysis was performed to relate individual associations and dissociations between the measures expressive vocabulary, cognition - measured by SON-R raw score - and MLU to the results of the profile group comparisons. In the individual assignments (*cf.* Figure 2) one can see that individuals with abilities < median are more likely to be assigned to the MN group, whereas individuals that show abilities ≥median are more likely to be assigned to the DN group. Despite repeated indications in previous analyses on the current sample that the vocabulary of the participants might be decisive for narrative abilities (see Neitzel and Penke, 2022b; Neitzel, under review), none of the three included variables could be identified as salient in the median split analyses and exact Fisher tests.

The described profiles point to implications for clinical work with the respective participants. On the one hand, it shows that different individuals with Down syndrome can reach a very variable level of (narrative) performance in adolescence. The participants in group DN might have an even higher narrative potential, which could be supported by addressing individual communicative strengths – nevertheless, since no longitudinal data is available in this investigation, this cannot be verified. This also has important clinical implications for the MN group, who may appear to have low levels of language or narrative skills, as AAC methods could be used more intensively in speech and language therapy with these participants rather than focusing solely on spoken language. In this context, it is a limitation of the present research that for the narrative analysis, only spoken language was included in the transcripts. For the future, it would be desirable to focus also on the non-verbal communication of the participants, for example pragmatic skills or gestural communication, and to investigate whether possible gesture usage might add supplementary narrative content to the children's and adolescents' output.

4.3. Limitations

The current study provides novel insights into single case-profiles of individuals with Down syndrome with regard to their narrative abilities, which may be transferable to other domains of language and cognition. At the same time, however, the study also underlies some limitations.

The described profile groups reflect the individual narrative abilities of children and adolescents with Down syndrome. Since corresponding comparison profiles of individuals from other clinical populations – e.g. individuals with (mixed) other intellectual disabilities – are not available, no statement can be made at this point as to whether these profiles are syndrome-specific for Down syndrome. A generalizability of the results is therefore not given, however, due to the selected individual case-oriented approach also not necessarily the goal of the study. The profile classifications shown are intended as a suggestion for the case-by-case classification of narrative performance and do not represent fixed categories.

Although the investigated sample of 28 children and adolescents with Down syndrome is of good size compared with other studies involving this population, the number of participants <30 individuals represents a statistical limitation. The formation of profile groups from this sample results in small numbers of participants representing each narrative profile (< 20 persons). It would be desirable to conduct similar analyses with a large number of participants, ideally >60 persons, to investigate whether tenable and statistically distinguishable profile groups can be verified for a larger group of individuals. At last, the sample studied shows little overall variability in terms of socioeconomic status and ethnicity. This should be taken into account in the recruitment of future samples. Another methodological limitation is that the MLU was calculated using the narrative transcripts presented here. Due to the already proven use of this material with participants with Down syndrome, a significant over- or underestimation of the utterance length is not to be assumed, but this cannot be completely excluded.

5. Conclusion and future research

The present study provides insights into the narrative abilities of children and adolescents with Down syndrome. The novel approach used here was to characterize individual narrative profiles, created on the basis of the NSS, beyond the performance of the whole group. It was found that the defined profile groups differed not only in terms of their narrative ability, but also in their general linguistic-cognitive profile. This allows the conclusion that narrative abilities could possibly be considered representative of the further linguistic-cognitive performance profile; however, further research in this area is necessary to draw firm conclusions. In this regard, the present analyses should only represent a starting point to conduct further investigations of individual narrative profiles and to explore individual developmental potentials, especially in the context of intervention studies. The aim of the present study was to provide a narrative profile per case, but not to examine individual aspects of macro- and microstructure in participants with Down syndrome in depth. However, as the data of the present article are fully available in open access, it is an intention of the author to initiate further research.

Data availability statement

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation. The full transcripts of the participants are available at CHILDES database: https://childes.talkbank.org/access/Frogs/German-Neitzel.html.

Ethics statement

The studies involving humans were approved by Ethics Committee of the Medical Faculty at University of Cologne. The studies were conducted in accordance with the local legislation and institutional requirements. Written informed consent for participation in this study was provided by the participants' legal guardians/next of kin.

Author contributions

IN collected the data, developed the analytical concept for the project, including the analyses presented in this manuscript, computed the all results, and wrote the manuscript.

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Conflict of interest

The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2023.1116567/full#supplementary-material

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Stuttering in individuals with Down syndrome: a systematic review of earlier research

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The main objective of this systematic review was to synthesize the evidence on the occurrence and characteristics of stuttering in individuals with Down syndrome and thus contribute knowledge about stuttering in this population. Group studies reporting outcome measures of stuttering were included. Studies with participants who were preselected based on their fluency status were excluded. We searched the Eric, PsychInfo, Medline, Scopus, and Web of Science Core Collection databases on 3rd January 2022 and conducted supplementary searches of the reference lists of previous reviews and the studies included in the current review, as well as relevant speech and language journals. The included studies were coded in terms of information concerning sample characteristics, measurement approaches, and stuttering-related outcomes. The appraisal tool for cross-sectional studies (AXIS) was used to evaluate study quality. We identified 14 eligible studies, with a total of 1,833 participants (mean = 131.29, standard deviation = 227.85, median = 45.5) between 3 and 58 years of age. The estimated occurrence of stuttering ranged from 2.38 to 56%, which is substantially higher than the estimated prevalence (1%) of stuttering in the general population. The results also showed that stuttering severity most often was judged to be mild-to-moderate and that individuals with Down syndrome displayed secondary behaviors when these were measured. However, little attention has been paid to investigating the potential adverse effects of stuttering for individuals with Down syndrome. We judged the quality of the evidence to be moderate-to-low. The negative evaluation was mostly due to sampling limitations that decreased the representability and generalizability of the results. Based on the high occurrence of stuttering and the potential negative effects of this condition, individuals with Down syndrome who show signs of stuttering should be referred to a speech and language pathologist for an evaluation of their need for stuttering treatment.

KEYWORDS

Down syndrome, stuttering, speech, disfluency, stuttering assessment, systematic review

1 Introduction

Stuttering is a speech-fluency disorder that involves the frequent and significant interruption of typical fluency and flow of speech, which can have negative effects on emotional, behavioral, and cognitive functioning from an early age (see, e.g., Craig et al., 2009; Briley et al., 2019; Guttormsen et al., 2021). One group that is reported to have a high occurrence of stuttering is individuals with Down syndrome (see, e.g., Kent and Vorperian, 2013). Due to language disorder (see, e.g., Martin et al., 2009; Næss et al., 2011), speech-sound disorder, and inappropriate prosody, speaking rate, and voice (see, e.g., Kent and Vorperian, 2013; Jones et al., 2019; Wilson et al., 2019; Loveall et al., 2021), an individual with Down syndrome typically have pervasive communication difficulties. Because stuttering may further interrupt their communication (see, e.g., Evans, 1977; Maessen et al., 2022), the identification of stuttering in this population is important in understanding the magnitude of their communication difficulties and supporting their communicative success.

Traditionally, in typically developing individuals, stuttering has mainly been operationalized and assessed based on behavioral factors (see, e.g., Tichenor and Yaruss, 2019). Examples of these factors include the type and number of disfluencies produced: audible symptoms that cause interruptions of speech, including repetitions of sounds (c-c-c-cat), syllables (ba-ba-ba-balloon), and one-syllable words (go-go-go); the prolongation of sounds (mmmilk); and blockages or stoppages of sounds (≠balloon). These audible symptoms are often accompanied by secondary behaviors caused by tension or the struggle to speak (i.e., visual symptoms, such as facial grimaces, blinking, or head nodding in an attempt to avoid stuttering; see, e.g., Bloodstein et al., 2021). Although there seems to be an agreement that speech behaviors are identifiers of stuttering, there is disagreement concerning which behaviors are symptomatic of stuttering, leading to differing operationalizations across studies (see, e.g., Einarsdottir and Ingham, 2005). One disagreement concerns whether word repetition is considered a stuttering disfluency. For example, in their operationalization of stuttering, Druker et al. (2020) excluded word repetitions, Millard et al. (2018) included word repetitions, and Boey et al. (2007) included only one-syllable word repetitions. Additionally, there have been various practices concerning the threshold at which speech disfluency is considered to be stuttering and, therefore, requires treatment. In a systematic review of stuttering-treatment studies by Sjøstrand et al. (2021), the frequency criterion (cutoff score) at treatment intake varied from no cutoff (Lewis et al., 2008) to a cutoff of a minimum of 3% of syllables stuttered (Harris et al., 2002; Lattermann et al., 2008). The use of frequency cutoff scores in the assessment of stuttering has been debated because (a) variability in stuttering across time and situations may cause participants to be wrongly classified if judgments are based on the percentage of stuttered syllables in only one speech sample (Constantino et al., 2016; Tichenor and Yaruss, 2021), (b) participants whose stuttering frequency is at the margins of the criterion set can be wrongly classified as non-stuttering (Tumanova et al., 2014), and (c) the adverse effects are not determined based on the frequency of overt speech disruptions (Koedoot et al., 2011; Blumgart et al., 2012), as the potential adverse effects of stuttering may also be critical for individuals with mild stuttering (i.e., mild based on listener evaluation; Beilby, 2014).

An increased awareness of the potential adverse effects of stuttering has led to a heightened focus on affective and cognitive reactions in the operationalization and assessment of stuttering. Affective reactions refer to feelings and emotions (e.g., feeling embarrassed, ashamed, or anxious), while cognitive reactions refer to a person's thoughts (e.g., anticipation) and identity (e.g., low self-confidence or self-esteem; Tichenor and Yaruss, 2019). Assessment procedures that are solely based on listener evaluations of observable behaviors can therefore be criticized for not considering the multidimensionality of stuttering. Based on a multidimensional understanding, the stuttering assessment will preferably also involve an evaluation made by the individual who stutters. As stuttering behavior can be highly variable across time and contexts (Tichenor and Yaruss, 2021), a combination of assessment approaches and outcome measures may provide a holistic picture of the condition. Additionally, for individuals with Down syndrome, who often have limited expressive language skills and short verbal expressions (see, e.g., Berglund et al., 2001; Chapman and Hesketh, 2001; Zampini and D'Odorico, 2011), it may be a challenge to record speech samples of at least 200 words, which is typically recommended for speech evaluation (Ward, 2018). Thus, using a combination of assessment strategies and outcome measures seems especially important for this clinical group.

Several narrative reviews of research on stuttering in individuals with Down syndrome exist (Zisk and Bailer, 1967; Stansfield, 1988; Van Borsel and Tetnowski, 2007; Kent and Vorperian, 2013; Bloodstein et al., 2021). These reviews refer to disagreements in the field concerning whether individuals with Down syndrome display genuine stuttering. These arguments are related to the simultaneous presence of other speech and communication disorders, as well as a lack of evidence for these individuals' secondary behaviors and awareness of their disfluency (Van Borsel and Tetnowski, 2007; Bloodstein et al., 2021). Challenges in the previous research literature have been highlighted. Operationalizations of stuttering are either not described or imprecisely described in several research reports (Zisk and Bailer, 1967; Kent and Vorperian, 2013), and the assessment procedures used in the typical population are not necessarily appropriate for individuals with disorders of intellectual development (Stansfield, 1988). Furthermore, several gaps in the research literature have been noted, such as limited knowledge concerning the presence or absence of secondary behaviors, the level of awareness and potential adverse effects of stuttering in this population (Zisk and Bailer, 1967; Van Borsel and Tetnowski, 2007), and whether stuttering is more common in male participants than female participants, as is suggested to be the case in the typical population (Van Borsel and Tetnowski, 2007). Additionally, Kent and Vorperian (2013) show a wide range in terms of participants' age within studies. As studies of stuttering in the typical population have found that both the occurrence (Reilly et al., 2009, 2013) and the overt and adverse symptoms of stuttering may change with age (Guitar, 2014), samples with wide age ranges may bias the results. These abovementioned reviews have not used a systematic approach (see, e.g., Higgins et al., 2022), do not cover the last decade of research in the field, and have a broad scope (e.g., focusing on speech impairment in general; Zisk and Bailer, 1967; Kent and Vorperian, 2013), and are, therefore, somewhat superficial in their review of the scientific stuttering research literature. The highlighted challenges and gaps in our knowledge about stuttering in individuals with Down syndrome call for an updated review of the literature, including a more in-depth discussion about how stuttering is operationalized and assessed in this clinical group. In the current review, we therefore summarize, assess, and synthesize the relevant existing research literature on stuttering in individuals with Down syndrome. Considering the potential negative effects of stuttering, a comprehensive overview of the relevant research has the potential to bolster the development of better strategies with which to identify those who stutter and may need treatment. The following research questions led the review process:

1. How is stuttering operationalized and measured in the included studies?
2. What is the estimated occurrence of stuttering in the included studies?
3. Does the estimated occurrence of stuttering in the included studies vary according to gender and age?
4. What characterizes stuttering in individuals with Down syndrome based on the findings of the included studies?

TABLE 1 Search strategy for PsychINFO.

Search strategy
1. (Down* syndrome or Trisomy 21 or mongol*).mp. [mp = title, abstract, heading word, table of contents, key concepts, original title, tests and measures, mesh]
2. (Stutter* or stammer* or disfluency* or non-fluency* or fluency disorder*).mp. [mp = title, abstract, heading word, table of contents, key concepts, original title, tests and measures, mesh]
3. 1 and 2

No restrictions were set with regard to publication date or language.

2 Methods

This article has been registered in Prospero in advance, and the registration ID is CRD42021273799.

To answer the research questions, we conducted a systematic literature review using explicit, accountable methods in line with standards prescribed by Gough and Thomas (2016) and Higgins et al. (2022). The study-selection process is presented using the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA; Page et al., 2021).

All statistical analyzes were conducted in SPSS statistics. We evaluated the strength of inter-rater agreement in the study-selection process, data extraction, and quality analysis by calculating Cohens's Kappa (κ; see, e.g., Gisev et al., 2013). Confidence intervals (95%) were calculated manually using the standard normal table (z-score table).

2.1 Eligibility criteria

In the current review, we included observational studies that reported at least one individual outcome measure of stuttering in individuals with Down syndrome. These could be studies that investigated stuttering via direct assessments or reports from a third party, such as parents or speech and language pathologists (SLPs). Studies in which the author(s) stated that they investigated stuttering were included. To answer the research questions, only studies that included occurrence estimates of stuttering (% and/or number) were eligible for inclusion. Thus, studies with samples that were preselected based on fluency status were excluded. Studies that investigated the co-existence of stuttering and other developmental speech disorders were included if the stuttering data were separated from other types of data. Mixed-etiology studies were considered for inclusion if they reported separate results for the participants with Down syndrome.

2.2 Search strategy

We developed the search strategy using words related to Down syndrome and stuttering. The search was conducted on 3rd January 2022 in the following databases: PsycINFO (Ovid interface, from 1806 onward), MEDLINE (Ovid interface, from 1946 onward), Eric (Ovid interface, from 1965 onward), Scopus (from 1960 onward), and Web of Science Core Collection (from 1945 onward). See Table 1 for the search strategy used in PsychINFO (Ovid interface, from 1806 onward). We verified and supplemented the electronic database search by searching (1) previous narrative reviews of stuttering or speech disfluency in individuals with Down syndrome and/or intellectual disability (Zisk and Bailer, 1967; Stansfield, 1988; Van Borsel and Tetnowski, 2007; Kent and Vorperian, 2013), (2) the reference lists of the included articles, and (3) acknowledged speech- and language-pathology journals and Google Scholar.

2.3 Study selection

The study-selection process had two phases. First, we screened the headings and abstracts and retrieved full-text sources that seemed to meet our inclusion criteria, as well as full-text sources that required further inspection. Second, we assessed the eligibility of these full-text sources. If a source appearing in our search was a chapter in an anthology, we read that specific chapter. When the source appearing in our search was a complete book, we first screened the index and then read the chapter(s) that were relevant to the topic of stuttering and/or disorders of intellectual development. See Appendix A for detailed information on the screening procedures.

2.3.1 Screening of headings and abstracts

The authors individually screened all titles and abstracts yielded by the systematic search against the eligibility criteria. Inter-rater reliability was calculated based on the agreement between the review authors regarding whether to include or exclude a study, as well as the reason for exclusion. There was good agreement between the review authors, $\kappa = 0.715$ (95% CI, 0.597 to 0.833), $p < 0.001$. Disagreements ($n = 18$ of 137 sources) were resolved through discussions between the review authors, including a reexamination of the headings and abstracts. In these discussions, the review authors were equal in status. Most disagreements concerned the reason for exclusion (i.e., not whether the study should be included). See Figure 1 (flow chart) for a record of the reasons for excluding sources in the heading- and abstract-screening phase.

2.3.2 Assessment of full-text documents

A total of 33 sources were sought for retrieval based on the systematic search. Of these 33 sources, we were able to retrieve full-text manuscripts from 32. Additionally, we sought full-text manuscripts of potentially relevant sources located through supplementary searches (previous reviews, the reference lists of included articles, and free searches in relevant journals and Google Scholar). The supplementary searches revealed 25 sources, Of these 25

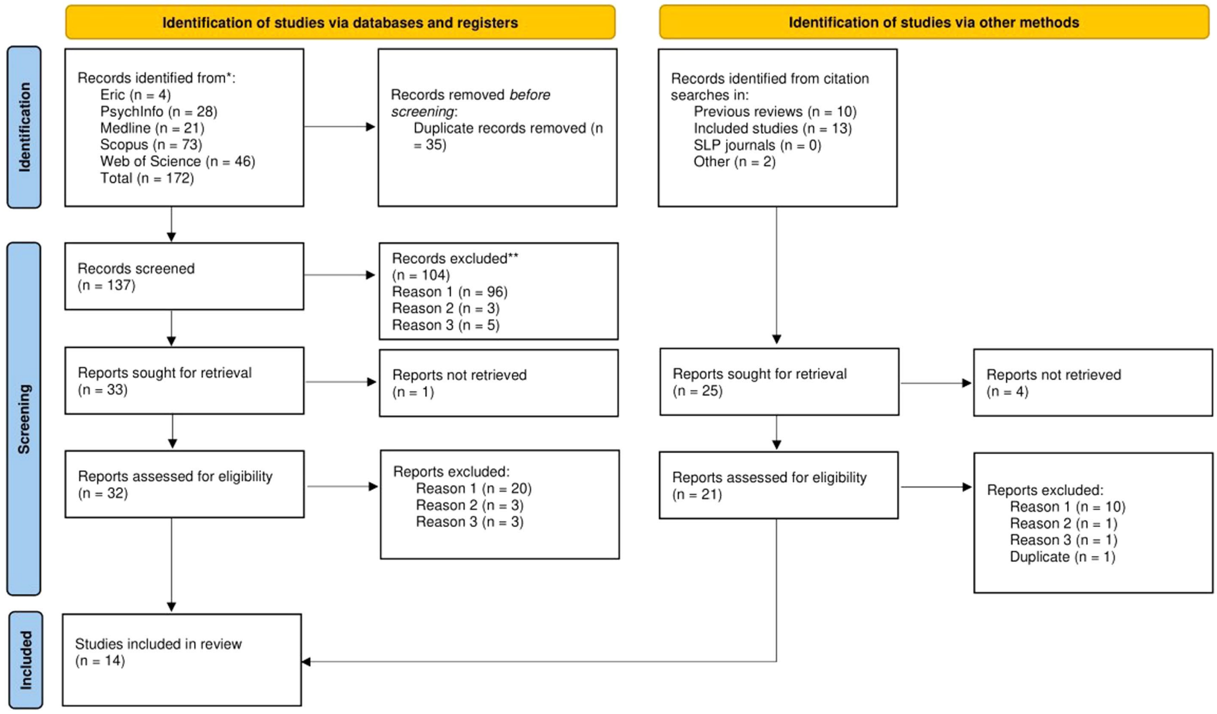

FIGURE 1
PRISMA flow diagram (Page et al., 2021).

sources we were able to retrieve full-text manuscripts from 21. This included our own study (Hokstad et al., 2022), which was not yet published at the time of the search. See the description of the data-extraction process and quality analysis for information about how this study was treated in the review process. One study was excluded without further assessment due to beeing a duplicate.

A total of 52 sources were assessed against the eligibility criteria in the full-text assessment phase. Three sources were assessed in collaboration between the review authors for training purposes (Evans, 1977; Wilcox, 1988; Borsel and Vandermeulen, 2008). During this training, we first assessed the sources independently against the eligibility criteria before comparing and discussing our decisions. We also revised our eligibility criteria when these sources were found to be ambiguous. Our own study, Hokstad et al. (2022), was assessed by an independent third party, a trained speech and language pathologist and assistant professor at the University of Oslo. Three sources were published in languages not mastered by the review authors (Kehrer, 1973; Rabensteiner, 1975; Takagi and Ito, 2007). These sources were assessed in collaboration with third party evaluators a trained speech and language pathologist and assistant professor at the University of Oslo whose first language is German and a professor at Nagoya University whose first language is Japanese.

The remaining 45 sources were screened individually and in duplicate. Evaluations were based on the agreement between the review authors regarding whether to include or exclude a study, as well as the reason for exclusion. There was good agreement between the review authors, $\kappa = 0.749$ (95% CI, 0.580 to 0.918), $p < 0.001$. Disagreements ($n = 7$ of 45 sources) were discussed and resolved between the two review authors, who were equal in status. Four of the disagreements concerned whether a source met the inclusion criteria. The remaining three disagreements concerned the reason for exclusion. The disagreements were resolved through a reexamination of the text and, on one occasion, making contact with the main author of one study for clarification (Maessen et al., 2021). See Figure 1 (flow chart) for a record of the reasons for excluding studies in the full-text-screening phase and Appendix B for examples of the characteristics of the excluded sources on topics related to stuttering in individuals with Down syndrome.

2.4 Data extraction process

A total of 14 studies were eligible for inclusion. We extracted information related to sample characteristics, measurement approaches, and outcomes. We developed the coding scheme for the data extraction and discussed the content of each category. Then, we selected four sources (Preus, 1972; Devenny and Silverman, 1990; Stansfield, 1990; Salihovic et al., 2012) that we collaboratively assessed to refine our coding categories and training before double-coding. The training included the independent assessment of each source based on our understanding of the coding scheme. Next, we compared our results and discussed our differences. In cases in which we found our category descriptions to be ambiguous, we revised these descriptions. See Appendix C for the coding scheme for data extraction. One source published in a language not mastered by the review authors was coded in collaboration with a third party (Rabensteiner, 1975), a trained speech and language pathologist and assistant professor at the University of Oslo, whose first language is German. The study by Hokstad et al. (2022) was coded by an independent coder who is a trained speech and language pathologist and assistant professor at the University of Oslo. For the remaining eight eligible studies, the review authors extracted data independently and in duplicate. Disagreements were resolved through a reevaluation of the text and discussions between the review authors, who were equal in status. We evaluated the strength of inter-rater agreement by calculating Cohen's kappa (κ) (see, e.g., Gisev et al., 2013) for each stuttering variable. The agreement between the review authors varied from good, $\kappa = 0.724$ (95% CI, 0.416 to 1.000), $p < 0.001$, to very good, $\kappa = 1.000$ (95% CI, 1.000, 1.000), $p < 0.001$. See Appendix D for κ values for each stuttering variable.

2.5 Quality appraisal

The appraisal tool for cross-sectional studies (AXIS) was used to assess the quality of the included studies. The tool includes items assessing sampling, justifications, clarity, and precision in descriptions of aims/objectives, methods, and results, as well as the reliability and validity of the measurement instruments (see Downes et al., 2016). For studies that included participants who did not have Down syndrome, we considered the information about the participants with Down syndrome only. In studies that investigated other areas of functioning, in addition to stuttering, we considered factors related to the measurement instruments and methodological transparency of the stuttering measures only (Q8–Q11). We made one adjustment when scoring the AXIS items; we only used the categories YES/NO (not using the "Do not know" category). Because scientific transparency is necessary for valid interpretations of the study results and the evaluation of research quality, negative evaluations were given when information in the study was lacking or insufficient for interpretation.

Again, one source (Rabensteiner, 1975) published in German, a language not mastered by the review authors, was coded in collaboration with a third party, and the authors' own study (Hokstad et al., 2022) was coded by an independent judge. Before coding and double-coding, the authors discussed each item of appraisal and selected three sources for training purposes (Preus, 1972; Devenny and Silverman, 1990; Salihovic et al., 2012). The authors coded the remaining nine studies independently and in duplicate. Disagreements were resolved through reassessments of the articles in question and discussions between the review authors, who were equal in status. We evaluated the strength of the inter-rater agreement for the quality assessment by calculating Cohen's kappa (see, e.g., Gisev et al., 2013). The inter-rater reliability was calculated based on the agreement between the review authors on each of the 20 AXIS items. Agreement varied from moderate, $\kappa = 0.630$ (95% CI, 0.297, 0.963), $p < 0.001$, to very good, $\kappa = 1.000$ (95% CI, 1.000, 1.000), $p < 0.001$. See Appendix E for the κ values for each AXIS item.

2.6 Data synthesis

We estimated the occurrence of stuttering in the total sample, per gender and per age group, by combining all samples included in the current review.

Occurrence = (the total number of individuals who stutter * 100)/ total N.

In the data synthesis, we used the occurrence estimates reported in each individual study, independent of how stuttering was operationalized, thus combining different operationalizations of stuttering. Furthermore, we evaluated sample characteristics, measurement approaches, and stuttering outcomes by conducting a narrative synthesis of the findings consisting of statistical (frequencies, numeric summarizations, average calculations, and numeric comparisons) and narrative (content comparisons and grouping in overarching categories) analyzes. The results are presented in text and table format.

3 Results

3.1 Study selection

A total of 14 studies met the eligibility criteria for inclusion in the current review. One study was the authors' own study (Hokstad et al., 2022), which was not yet published at the time of the systematic search. The remaining studies were identified through (1) a systematic search ($n = 6$), (2) a search of the reference lists of previous reviews ($n = 6$), and (3) the reference lists of included studies ($n = 1$). See Figure 1 for a flow chart depicting the selection process.

3.2 Study characteristics

The included studies were published between 1955 and 2022 and, as such, represent seven decades of research on stuttering in individuals with Down syndrome. However, most of the studies are older, with a majority ($n = 10$) having been published before 2000.

3.3 Operationalization of stuttering

Four studies did not contain any operationalization of stuttering (Gottsleben, 1955; Rabensteiner, 1975; Kumin, 1994; Schieve et al., 2009). In eight studies, stuttering was operationalized based on indicators related to speech behaviors alone (Rohovsky, 1965; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022) or in combination with secondary behaviors (Schlanger and Gottsleben, 1957; Martyn et al., 1969; Keane, 1970; Preus, 1972; Devenny and Silverman, 1990) and on affective and cognitive reactions to stuttering (Martyn et al., 1969; Keane, 1970; Preus, 1972). See Table 2 for an overview of the indicators included in the operationalization of stuttering across studies.

In addition to the presence of indicators of stuttering, four studies also reported the threshold at which (e.g., % syllables stuttered cutoff score) stuttering behaviors were considered clinically significant (Keane, 1970; Preus, 1972; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022). See Table 3 for an overview of the frequency

TABLE 2 Operationalization of stuttering.

Indicators	Studies													
	1	2	3	4	5	6	7	8	9	10	11	12	13	14
Speech behaviors[a]	–	–	–	–	–	–	–	–	–	–	✓	–	–	✓
Repetitions[b]	✓	–	–	–	–	–	✓	–	–	–	–	–	✓	–
Part-word repetitions[c]	–	✓	–	✓	✓	–	–	✓	–	✓	–	–	–	–
Single-syllable word repetitions	–	✓	–	✓	✓	–	–	✓	–	✓	–	–	–	–
Multisyllabic whole-word repetitions	–	–	–	–	–	–	–	✓	–	✓	–	–	–	–
Prolongations	✓	✓	–	✓	✓	–	✓	✓	–	✓	–	–	–	–
Blocks[d]	✓	✓	–	✓	✓	–	✓	–	–	✓	–	–	✓	–
Secondary behaviors	✓	–	–	–	✓	–	✓	✓	–	–	✓	–	✓	✓
Cognitive reactions	–	–	–	–	✓	–	✓	✓	–	–	–	–	–	–
Affective reactions	–	–	–	–	✓	–	✓	–	–	–	–	–	–	–

1 = Devenny and Silverman (1990), 2 = Eggers and Van Eerdenbrugh (2018), 3 = Gottsleben (1955), 4 = Hokstad et al. (2022), 5 = Keane (1970), 6 = Kumin (1994), 7 = Martyn et al. (1969), 8 = Preus (1972), 9 = Rabensteiner (1975), 10 = Rohovsky (1965), 11 = Salihovic et al. (2012), 12 = Schieve et al. (2009), 13 = Schlanger and Gottsleben (1957), 14 = Stansfield (1990). [a]Speech behaviors not specified. [b]Types of repetitions not specified. [c]Includes the repetition of sounds and the repetition of syllables. [d]Eggers and Van Eerdenbrugh (2018) report broken words (blocks within words) separately.

TABLE 3 Frequency cutoff scores.

Study	Cutoff	Unit of analysis
Eggers and Van Eerdenbrugh (2018)	3 or more	Per 100 syllables or a maximum number of syllables
Hokstad et al. (2022)	3 or more	Per total number of syllables
Keane (1970)	3 or more	Per total number of words
Preus (1972)	5 or more	Per 100 words

cutoff scores used in these four studies. Finally, two studies (Stansfield, 1990; Salihovic et al., 2012) operationalized stuttering based on the frequency of stuttering disfluencies, the duration of stuttering blocks, and the number of physical concomitants (i.e., secondary behaviors), without specifying which types of disfluencies were considered and at what threshold disfluencies were considered stuttering. See Appendix F for a detailed overview of the operationalizations of stuttering in the included studies.

3.4 Measurement approaches

In two studies, stuttering was assessed indirectly through parental reports (Kumin, 1994; Schieve et al., 2009). As the parents simply reported whether their child stuttered or not, these studies provided limited information about stuttering besides the stuttering occurrence estimate.

In 12 studies, stuttering was identified through clinical judgment by either SLPs/SLP students and/or the researcher(s) themselves (Gottsleben, 1955; Schlanger and Gottsleben, 1957; Rohovsky, 1965; Martyn et al., 1969; Keane, 1970; Preus, 1972; Rabensteiner, 1975; Devenny and Silverman, 1990; Stansfield, 1990; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022). In eight of these 12 studies, stuttering was identified through speech-sample analysis (Rohovsky, 1965; Keane, 1970; Preus, 1972; Devenny and Silverman, 1990; Stansfield, 1990; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022), while in four studies, stuttering was identified through either written sources (Gottsleben, 1955; Schlanger and Gottsleben, 1957) or real-time observation (Martyn et al., 1969; Rabensteiner, 1975). Spontaneous speech samples were commonly elicited through planned speaking situations, such as play sessions (Eggers and Van Eerdenbrugh, 2018), conversations about pictures (Preus, 1972; Salihovic et al., 2012), and story retelling (Rohovsky, 1965; Hokstad et al., 2022). In six studies, audio data were collected (Rohovsky, 1965; Preus, 1972; Stansfield, 1990; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022), while video data were collected in two studies (Keane, 1970; Devenny and Silverman, 1990). The length of the speech samples and the amount of speech material collected varied greatly across studies; however, the speech samples were, with one exception (Hokstad et al., 2022), retrieved from only one speaking situation. Furthermore, while some studies used speech samples with variable lengths and amounts of speech (Preus, 1972; Hokstad et al., 2022), others based their evaluation on a set speech-sample length (Rohovsky, 1965; Keane, 1970; Stansfield, 1990) or a set number of words or syllables (Preus, 1972; Devenny and Silverman, 1990; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018). See Table 4 for a detailed overview of the measurement approach(es) used in each study.

3.5 Sample characteristics

A total of 1,833 (*M* = 131.29, SD = 227.85, median = 45.5) individuals with Down syndrome participated in the included studies, with sample sizes ranging from 26 to 897 participants. Kumin (1994) represented an extreme value (± 3 standard deviations from the mean) with 897 participants. The average sample size with this extreme outlier excluded was 72 participants (SD = 60.46, min = 26, max = 200). Participants of all ages were represented across the included studies. The studies that reported participant age had wide age ranges (Gottsleben, 1955; Rohovsky, 1965; Keane, 1970; Preus, 1972; Rabensteiner, 1975; Devenny and Silverman, 1990; Schieve et al., 2009; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022), with an average age gap of 21 years between the youngest and oldest participants. Hokstad et al. (2022) had the narrowest age spread, at 2 years, while Gottsleben (1955) had the widest age spread, at 43 years. Eight studies reported the gender distribution of the participants (Gottsleben, 1955; Rohovsky, 1965; Keane, 1970; Preus, 1972; Devenny and Silverman, 1990; Schieve et al., 2009; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022), and a total of 310 male and 278 female participants took part in these studies. Seven studies reported including only individuals who used speech (Gottsleben, 1955; Rohovsky, 1965; Preus, 1972; Devenny and Silverman, 1990; Kumin, 1994; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022). However, the level of speech proficiency is often not specified. See Table 5 for a detailed overview of the sample characteristics.

3.6 The occurrence of stuttering in individuals with Down syndrome

The reported occurrence of stuttering varied between 2.38 and 56.00% across the included studies. Combining all samples (total number of individuals who stutter * 100/total N) resulted in an occurrence estimate of 19.80%. Occurrence by gender or the information necessary to calculate occurrence by gender was reported in five studies (Gottsleben, 1955; Rohovsky, 1965; Keane, 1970; Devenny and Silverman, 1990; Eggers and Van Eerdenbrugh, 2018). The results suggest a gender factor of 2:1; 27.11% (45 of 166) of the male participants and 13.64% (21 of 154) of the female participants in these studies were determined to stutter. Information about the age of the stuttering participants was reported in six studies, and their ages ranged from 5 to 58 years of age (Gottsleben, 1955; Rohovsky, 1965; Keane, 1970; Stansfield, 1990; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018). When categorizing the included studies based on age groups (preschool age, school age, adulthood, and mixed), we found seven studies reported on mixed-age samples: from preschool age through adulthood (Kumin, 1994), preschool age through school age (Schieve et al., 2009; Eggers and Van Eerdenbrugh, 2018), and school age through adulthood (Gottsleben, 1955; Keane, 1970; Preus, 1972; Stansfield, 1990). Three of these studies reported separate findings based on age or age intervals, but the participants were not equally distributed across age groups (Stansfield, 1990; Kumin, 1994; Eggers and Van Eerdenbrugh, 2018). Three studies reported on only school-aged participants (Rohovsky, 1965; Salihovic et al., 2012; Hokstad et al., 2022), while one study reported on only adults (Devenny and Silverman, 1990). The remaining three studies could not be categorized due to the lack of information on participant age (Schlanger and Gottsleben, 1957; Martyn et al., 1969; Rabensteiner, 1975). See Table 6 for detailed information on the occurrence of stuttering across studies. See Table 7 for an overview of the occurrence of stuttering across age groups.

3.7 Stuttering outcomes

Stuttering frequency in individuals who stuttered was reported in four studies (Keane, 1970; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022). The approaches to calculating stuttering frequency varied, and these studies were, therefore, not directly comparable. For example, while Eggers and Van Eerdenbrugh (2018) reported an average of 5.1% stuttering-like disfluencies per 100 syllables, Keane (1970) reported an average of 11.35% stuttered words. The distribution of disfluency types in individuals who stutter was only reported in the dissertation by Keane (1970). Based on her reporting of stuttering types, 78% of the disfluencies in individuals with Down syndrome who stuttered (*n* = 20) were prolongations, which occurred in 19 of 20 participants, and 22% were part-word repetitions, which occurred in 17 of 20 participants.

Stuttering severity was reported in six studies. In half of these studies (Stansfield, 1990; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018), judgments were based on the total score on the Stuttering Severity Instrument (Riley, 1980, 1994), while in the other half (Rohovsky, 1965; Keane, 1970; Hokstad et al., 2022), judgments were based on placement on a severity scale after a perceptual evaluation. According to the findings of these studies, most of the participants displayed mild-to-moderate stuttering, and severe stuttering was rare.

Secondary behaviors were reported in six studies (Rohovsky, 1965; Keane, 1970; Preus, 1972; Stansfield, 1990; Salihovic et al., 2012; Eggers and Van Eerdenbrugh, 2018). Four of these studies reported the number of stuttering participants displaying secondary behaviors (Rohovsky, 1965; Keane, 1970; Stansfield, 1990; Eggers and Van Eerdenbrugh, 2018), while for the remaining two studies, this information was not reported (Salihovic et al., 2012) or was unclear (Preus, 1972). Across the studies that did report the occurrence of secondary behaviors, these behaviors were observed in 66.13% of the stuttering participants.

Outcomes related to the potential adverse effects of stuttering were reported in three studies (Martyn et al., 1969; Keane, 1970; Preus, 1972), all of which concluded that there was no evidence of affective or cognitive reactions in their participants. However, it must be noted that this is our interpretation based on the descriptive information that exists in these research reports. Martyn et al. (1969) stated that stuttering, in their participants, did not appear to be associated with anticipation or avoidance, while Preus (1972) stated that, even though there were signs of avoidance and postponement in some participants, none of them seemed to be aware of or embarrassed by their stuttering.

TABLE 4 Measurement approaches.

Study	Assessor(s)	Instrument		
	Parent(s), SLP(s), author(s)/researcher(s), student(s), stutterer, or other	Clinical judgment, parental judgment, self-report, or other	Speaking situation as described in study	Speech sample (audio/video, duration and number of utterances, words, or syllables), written sources, real-time observation (duration and/or number of utterances, words, or syllables), or own experience
Devenny and Silverman (1990)	SLPs	Clinical judgment	Conversation about work and recreation	Speech sample (video, 10 min, first 150 words)
Eggers and Van Eerdenbrugh (2018)	Authors	Clinical judgment	Play session with toy or book adapted to age and interests	Speech sample (audio, 15 min, 50 utterances[1])
Gottsleben (1955)	Author and SLPs	Clinical judgment	NR	Written sources
Hokstad et al. (2022)	Researchers	Clinical judgment	Picture book dialog and story-retelling	Speech sample (audio, unknown duration/number of utterances/words/syllables)
Keane (1970)	1 = SLP 2 = SLPs and other (clinical experience with stutterers)	Clinical judgment	1 and 2 = Interviews about daily life and interests	1 = Real-time observation (*ca.* 10 min), 2 = Speech sample (video, mean duration 10 min)
Kumin (1994)	Parent	Parental judgment	NR	Real-time observation (NR)
Martyn et al. (1969)	SLPs and students	Clinical judgment	Conversation, interview, or reading sample adapted to the level of intellectual disability	Real-time observation (NR)
Rohovsky (1965)	10 grad. Students (speech and hearing science)	Clinical judgment	Story retelling	Speech sample (audio, 30 s)
Salihovic et al. (2012)	SLPs	Clinical judgment	Spontaneous speech elicited through pictures	1 and 2 = Speech sample (audio, minimum 200 syllables) 3 = real-time observation (minimum 200 syllables)
Schieve et al. (2009)	Adult family member (usually parent)	Parental judgment	NR	Real-time observation (NR)
Schlanger and Gottsleben (1957)	Authors/researchers	Clinical judgment	NR	Written sources
Stansfield (1990)	1 = Other (nursing or ATC staff) 2 and 3 = SLP and students	1 = Other (paid caregivers) 2 and 3 = Clinical judgment	1 = NR 2 = Informal interaction 3 = Assessment situation	1 = Real-time observation (NR) 2 = Real-time observation (5 min) 3 = Speech sample (audio, 30 min)
Preus (1972)	(1) NR (2) Other (personnel day institutions) (3) NR	Clinical judgment	(1) Spontaneous speech evoked by means of conversation pictures (2) Daily interaction (3) NR	(1) Speech sample (audio, mean duration 9.47 min, min/max = 3.5–28 min, minimum 200 words) (2) Real-time observation (NR) (3) NR

In cases where assessments have been conducted in several stages, each stage is numbered, SLP, speech and language pathologist; min, minutes; NR, not reported. [1] When 50 utterances were not available, the maximum number of utterances was used. In cases in which assessments have been conducted in several stages, each stage is numbered, SLP, speech and language pathologist; ATC staff, adult training center staff; min, minutes; NR, not reported; grad, graduate.

Finally, Keane (1970) placed all stuttering individuals in one of Bloodstein's four developmental phases of stuttering (Bloodstein, 1960) and reported that none of the participants had reached phase four, advanced stuttering, which includes the anticipation of stuttering, word substitutions, and avoidance of speaking, as well as evidence of fear and embarrassment. None of the more recent studies included

TABLE 5 Sample characteristics.

Study	Nationality	Sample size (N)	Gender		Age			Language proficiency
			Male	Female	M	Min	Max	
Devenny and Silverman (1990)	United States	31	20	11	41	30	58	Used speech[1]
Eggers and Van Eerdenbrugh (2018)	Belgium	26	12	14	8	3	12	Used speech[2]
Gottsleben (1955)	United States	36	23	13	27	9	52	Used speech[2]
Hokstad et al. (2022)	Norway	75	40	35	7	6	8	Used speech[2]
Keane (1970)	United States	200	100	100	15	6	46	Participants did and did not use speech[2]
Kumin (1994)	United States	897	NR	NR	NR	NR	NR	Used speech[2]
Martyn et al. (1969)	United States	42	NR	NR	NR	NR	NR	NR
Preus (1972)	Norway	47	21	26	NR	7	48	Speech intelligibility[3]
Rabensteiner (1975)	Germany	49	NR	NR	NR	5	15	Good receptive language skills[2]
Rohovsky (1965)	United States	27	11	16	15	9	20	Used three-word utterances or more
Salihovic et al. (2012)	Bosnia-Herzegovina	37	NR	NR	NR	6	17	NR
Schieve et al. (2009)	United States	146	83	63	NR	3	17	NR
Schlanger and Gottsleben (1957)	United States	44	NR	NR	NR	NR	NR	All but one participant used speech[2]
Stansfield (1990)	Scotland	176	NR	NR	NR	NR	NR	NR

NR, not reported; Min, minimum; Max, maximum. [1]Indicated by verbal IQ of 43–77. [2]As stated in the study. [3]Judges able to understand half of the responses.

information on affective or cognitive reactions to stuttering. See Table 8 for the stuttering outcomes and information regarding the instruments these outcomes are based on.

3.8 Quality of the included studies

3.8.1 Inter-rater reliability

Inter-rater reliability was given for some or all stuttering measures reported in three of the included studies (Keane, 1970; Eggers and Van Eerdenbrugh, 2018; Hokstad et al., 2022). See Table 9 for an overview of inter-rater reliability measures. Based on the low number of stuttering outcome measures tested for consistency across studies, test validity is an area of great insecurity in existing research on stuttering in individuals with Down syndrome. Additionally, research on typically developing individuals has raised concerns about the inter-rater reliability of both the Stuttering Severity Instrument (SSI; Davidow, 2021) and disfluency-type measures (Cordes, 2000; Einarsdottir and Ingham, 2005), both of which have been used across studies in the current review. It is therefore important, especially with this population, which has profound speech and language difficulties (see, e.g., Martin et al., 2009; Næss et al., 2011; Jones et al., 2019; Wilson et al., 2019; Loveall et al., 2021), that speech evaluations are performed by more than one rater.

3.8.2 Study quality appraisal

The studies in the current review met between 6 and 19 of 20 potential criteria items of the AXIS tool ($M = 11.07$, SD = 3.99). Thus, the results of the quality appraisal indicate that the quality of the included studies ranges from low to high, with most studies being of moderate-to-low quality. Many studies were found to be lacking in areas related to sampling procedures. First, most studies had small sample sizes, and all studies lacked sample size justifications (AXIS item 3). To provide an example regarding the number of participants necessary for an accurate estimation of stuttering frequency in a population, given an estimated population of 3,725 individuals with Down syndrome in Norway (De Graaf et al., 2021), the minimum sample size for determining the frequency of stuttering in the Norwegian population is 349 participants (95% confidence level, anticipated frequency unknown; Dean et al., 2013). Additionally, as the incidence of stuttering is known to be influenced by the age group sampled (Reilly et al., 2009, 2013), the appropriate sample size is likely to be even higher. Second, convenience sampling was common across studies, and only two studies reported systematic recruitment procedures (Schieve et al., 2009; Hokstad et al., 2022). Thus, for most of the included studies, it was unlikely that samples closely represented the population of individuals with Down syndrome they were drawn from (AXIS items 5 and 6). Small samples recruited through convenience sampling are not well suited to providing estimates of

TABLE 6 Occurence of stuttering.

Study	Occurrence		Occurrence male/female		Occurrence by age group	Age of stutterers	
	%	*n*	%	*n*		*M*	Min/Max
Devenny and Silverman (1990)	42.00	13	45.00/36.00	9/4	Adults 13/31	NR	NR
Eggers and Van Eerdenbrugh (2018)	31.00	8	50.00/14.00	6/2	Pre-schoolers 1/4 School-age 7/22	10	5/13
Gottsleben (1955)	33.00	12	44.00/15.00	10/2	Mixed 12/36	NR	16/43
Hokstad et al. (2022)	53.34	40	NR	NR	School-age 40/75	NR	NR
Keane (1970)	10.00	20	16.00/4.00	16/4	Mixed 20/200	18	6/32
Kumin (1994)	17.00	153	NR	NR	Pre-schoolers 4/191 School-age 115/561 Adults 34/145	NR	NR
Martyn et al. (1969)	2.38	1	NR	NR	NR	NR	NR
Preus (1972)	40.95	19	NR	NR	Mixed 14/47	NR	NR
Rabensteiner (1975)	33.33	11	NR	10/1	NR	NR	NR
Rohovsky (1965)	48.00	13	36.36/56.25	4/9	School-age 13/27	15	9/19
Salihovic et al. (2012)	13.51	5	NR	NR	School-age 5/37	14	10/17
Schieve et al. (2009)	15.60	27	NR	NR	Mixed 27/146	NR	NR
Schlanger and Gottsleben (1957)	45.45	20	NR	NR	NR	NR	NR
Stansfield (1990)	11.93	21	NR	13/8	School-age: 1/NR Adults: 19/NR Missing: 1[1]	33	17/61

[1] In Stansfield (1990), detailed information about one of the stuttering participants with Down syndrome is missing.

TABLE 7 Occurrence by age group.

Age group	Studies	Participants	Occurrence of stuttering	
			N	%
Mixed samples	4	429	78	18.01
Adulthood	2	176	47	26.70
School age	6	771	191	24.77
Preschool age	2	195	5	2.56
NR	3			

N = number of participants. Studies that reported separate findings based on age or age intervals (Stansfield, 1990; Kumin, 1994; Eggers and Van Eerdenbrugh, 2018) are represented within more than one category. Each participant is represented once.

occurrences, as random sampling and adequate sample sizes are necessary for precise prevalence and incidence estimates (Munn et al., 2014). Inaccuracies in the occurrence estimates and related outcomes must therefore be assumed. A related area of concern is the treatment and reporting of non-responders (AXIS items 7, 13, and 14), which is unreported or unclear in several studies. Given the large variation in speech and language proficiency in this population (Karmiloff-Smith et al., 2016), it is, for example, likely that some participants across studies did not provide sufficient speech and/or intelligible speech for an evaluation of stuttering to be conducted. However, in many of the included studies, it is generally not clear whether and how many participants were lost due to restricted speech and language. See Table 10 for an overview of the quality assessment of the included studies for each AXIS item.

4 Discussion

The current review has five main findings: (1) there was no common approach to identifying stuttering in individuals with Down syndrome, but there was a one-sided focus on observational aspects; (2) the occurrence estimates were generally high but varied across studies; (3) the occurrence estimates were higher in school-aged and adult groups than in the preschool-aged group; (4) the occurrence estimates were higher in male than in female participants; and (5) stuttering was mild-to-moderate, and secondary behaviors were found when measured.

To identify stuttering, the included studies used various assessment approaches, which were initially developed for the typically developing population, mainly focusing on the identification of speech disfluencies, both with and without frequency cutoff scores. However, no studies included self-reports of experiences related to stuttering. One reason for the heavy focus on stuttering behavior may be the high frequency of older studies included in this review. The multidimensional view of stuttering may represent a more recent understanding of the disorder, one in line with the changes in the diagnostic criteria for stuttering in the latest revision of the International Classification of Diseases (ICD-11, World Health

TABLE 8 Stuttering outcomes.

Study	Stuttering behavior				Adverse impacts	
	Total frequency	Frequency per disfluency type	Severity	Secondary behavior	Affective reactions	Cognitive reactions
Devenny and Silverman (1990)	NR	NR	NR	NR	NR	NR
Eggers and Van Eerdenbrugh (2018)	%SLD[a] M 5.1 min/max 3–11 SSI score[b] M 10	NR	n per category [c] Mild 5, Moderate 3 SSI score [c] M 18.38 min/max 12–26	Participants[d] 6 of 8 SSI score[d] M 3.8	NR	NR
Gottsleben (1955)	NR	NR	NR	NR	NR	NR
Hokstad et al. (2022)	%SLD [e] M 9.50 min/max 3.22–29.37	NR	Severity rating[f] M 2.30 min/max 0.50–6.50	NR	NR	NR
Keane (1970)	%SLD[g] per minute M 7.51 total M 11.35	Relative frequency part-word repetitions 22% prolongations 78% Participants with part-word repetitions 17/20 prolongations 19/20	n per category Very mild/Mild 1, Mild 2, Mild/Moderate 10, Moderate 5, Moderate/Severe 2	Participants 18 of 20	No affective reactions[h]	No cognitive reactions[h]
Kumin (1994)	NR	NR	NR	NR	NR	NR
Martyn et al. (1969)	NR	NR	NR	NR	NR	No cognitive reactions
Preus (1972)	NR	NR	NR	NR*	No affective reactions	No cognitive reactions
Rabensteiner (1975)	NR	NR	NR	NR	NR	NR
Rohovsky (1965)	NR	NR	n per category[a] Mild 7, Moderate 6 Severity rating[a] M = 1.5 Min/max = NR	Participants 5 of 13	NR	NR
Salihovic et al. (2012)	SSI score M 11.60 min/max 8–16	NR	SSI score M 26[b] min/max 17–49	SSI score M 6.40 min/max 3–12	NR	NR
Schieve et al. (2009)	NR	NR	NR	NR	NR	NR
Schlanger and Gottsleben (1957)	NR	NR	NR	NR	NR	NR
Stansfield (1990)	NR	NR	n per category Mild 7, Moderate 7, Severe 6, Missing 1	SSI score NR Participants 12 of 21 Missing 1[c]	NR	NR

%SLD, percentage of syllables stuttered; SSI, stuttering severity instrument (Riley, 1994). [a]Percent syllables stuttered per 100 syllables or maximum number of syllables when 100 syllables were not available. [b]Based on the frequency score from the SSI (Riley, 1994). [c]Based on the total score from the SSI (Riley, 1994). [d]Based on the physical concomitant score from the SSI (Riley, 1994). [e]Percentage of syllables stuttered per maximum number of syllables in each speech sample. [f]Based on the stuttering severity rating (0–9) on the Stuttering Severity Rating Scale (Onslow et al., 2020). [g]Percentage of stuttered words. [h]Based on placement in one of Bloodstein's four developmental stages of stuttering (Bloodstein, 1960). SSI, stuttering severity instrument (Riley, 1980, 1994). [a]Based on stuttering severity rating on a scale from 0 to 5. [b]Equals moderate degree of stuttering. [c]In Stansfield (1990), detailed information about one of the stuttering participants with Down syndrome is missing.

Organization, 2022), to include the significant effects on functioning (e.g., social communication and personal and family life) in addition to observable behaviors. Another reason for the heavy focus on stuttering behavior may be low expectations regarding these individuals' capability to evaluate and report their own reactions due to reduced language skills (Martin et al., 2009; Næss et al., 2011) and

TABLE 9 Inter-rater reliability.

Study	Variable	Reliability
Devenny and Silverman (1990)	Occurrence	NR
Eggers and Van Eerdenbrugh (2018)	%SLD[a] SSI frequency score SSI physical Concomitant score SSI Total score (severity)	Agreement index percentage = 0.91[b] NR NR NR NR
Gottsleben (1955)	Occurrence	NR
Hokstad et al. (2022)	%SLD[a] Severity rating[a]	Percent agreement = 89.07 Percent agreement = 93.75[c]
Keane (1970)	%SLD[a] Severity rating Secondary reactions Affective reactions Cognitive reactions	Pearson's product moment Correlation coefficient = 0.970 NR NR NR NR
Kumin (1994)	Occurrence	NR
Martyn et al. (1969)	Occurrence Cognitive reactions	NR NR
Preus (1972)	Occurrence	NR
Rabensteiner (1975)	Occurrence	NR
Rohovsky (1965)	Occurrence Severity rating Secondary behavior	NR NR NR
Salihovic et al. (2012)	SSI total score[a] SSI frequency score SSI physical Concomitant score	NR NR NR
Schieve et al. (2009)	Occurrence	NR
Schlanger and Gottsleben (1957)	Occurrence	NR
Stansfield (1990)	SSI total score[a] SSI physical Concomitant score	NR NR

[a] Basis for occurrence estimate. [b] 'Agreement index' percentage (i.e., the number of agreements divided by the sum of agreements and disagreements regarding all disfluencies). [c] Scores that were within one scale point of one another were judged to indicate agreement between the raters.

reduced non-verbal mental ability (Næss et al., 2021). However, reactions to stuttering are found in typically developing children from a very low chronological age (Boey et al., 2009), which may correspond to the lower developmental age in individuals with Down syndrome. Furthermore, the existence of affective and cognitive reactions to stuttering has been described in both children (Bray, 2017) and adults (Jackson et al., 2014) with Down syndrome. Based on these indications, individuals with Down syndrome's own evaluations can be considered in the identification of stuttering.

Although the results of this review showed that listener evaluation in the form of clinical judgment was a common approach, the factors considered to be indicative of stuttering varied across studies, especially those related to types of repetition, as did what threshold disfluencies were considered clinically significant. However, research on typically developing individuals faces the same challenges, which means that comparisons are restricted between both the studies in this review and research projects on stuttering in general (Einarsdottir and Ingham, 2005). The lack of agreement in the field about the indicators of stuttering, as well as at what threshold (cut-off) disfluency is considered stuttering may influence who is considered to need treatment. It may also fuel the discussion about whether the disfluencies seen in individuals with Down syndrome represent genuine stuttering.

In addition to these general challenges within the field of stuttering related to assessment, there are some specific challenges related to the identification of stuttering behaviors in individuals with Down syndrome. As language development is significantly delayed in this population, there are likely differences in the amount of speech material available for listener evaluation across age groups; preschool-aged children with Down syndrome who have begun to speak will, for example, often produce short utterances (Berglund et al., 2001; Zampini and D'Odorico, 2011). This means that the amount of speech material elicited in one speaking situation may be very limited for some participants, as is the case in, e.g., Eggers and Van Eerdenbrugh (2018). As previously mentioned, concerns have been raised regarding the poor reliability of stuttering measures based on the identification of speech disfluencies in speech samples (Cordes, 2000; Einarsdottir and Ingham, 2005; Davidow, 2021), suggesting that the identification of stuttered disfluencies in the typical population can be challenging. Limited speech, in combination with atypical speech features in individuals with Down syndrome, may pose an added challenge in this regard. The reviewed studies that did include inter-rater reliability analysis did, however, report good or very good reliability. The same results were found in Maessen et al. (2023), who included a preselected sample of individuals with Down syndrome who stuttered. Whether the good reliability scores in these studies are related to, for example, speech characteristics, including the frequency of stuttering types, or to the use of summary agreement scores across all disfluency types (i.e., each disagreement has less influence when the number of stuttering disfluencies is large) is unknown.

The abovementioned concerns and limitations related to stuttering assessment, in addition to the quality of the included studies, constitute the frame within which we can interpret and understand the findings of the current review. Nevertheless, studies consistently report high occurrences of stuttering in individuals with Down syndrome when interpreted against the estimated 1% prevalence for the typical adult population (see, e.g., Månsson, 2000) and the 5–11% cumulative incidence of stuttering in typically developing children (Reilly et al., 2009, 2013). The fact that a high percentage of the individuals in the included studies displayed core stuttering behaviors to a degree which they were judged to stutter is in line with previous research, showing that individuals with Down syndrome are vulnerable to speech, language, and communication difficulties (see, e.g., Martin et al., 2009; Næss et al., 2011; Jones et al., 2019; Wilson et al., 2019; Loveall et al., 2021), including stuttering (Kent and Vorperian, 2013). Recent research has also shown that coexisting speech and/or language disorders are common in individuals who stutter and do not have Down syndrome (Wolk and LaSalle, 2023), as are comorbidities between stuttering and other neurodevelopmental disorders (e.g., disorders of intellectual development; Briley and Ellis,

TABLE 10 Quality appraisal.

Study	AXIS item																				Total score per study
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	
Devenny and Silverman (1990)	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟥	🟥	🟩	🟥	🟥	🟥	🟥	🟩	🟥	🟩	🟥	🟩	🟥	7
Eggers and Van Eerdenbrugh (2018)	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟩	🟩	🟩	🟩	🟥	🟥	🟥	🟩	🟩	🟩	🟩	🟩	🟩	13
Gottsleben (1955)	🟩	🟩	🟥	🟩	🟥	🟥	🟩	🟩	🟥	🟩	🟥	🟩	🟩	🟩	🟩	🟥	🟩	🟥	🟩	🟥	12
Hokstad et al. (2022)	🟩	🟩	🟥	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	19
Keane (1970)	🟩	🟥	🟥	🟩	🟥	🟥	🟩	🟩	🟩	🟩	🟩	🟩	🟥	🟩	🟩	🟩	🟩	🟩	🟩	🟥	14
Kumin (1994)	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟩	🟥	🟩	🟥	🟥	🟥	🟥	🟥	🟥	🟥	🟥	🟩	🟥	6
Martyn et al. (1969)	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟩	🟩	🟩	🟩	🟥	🟥	🟥	🟩	🟥	🟩	🟥	🟩	🟥	10
Preus (1972)	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟩	🟩	🟩	🟥	🟥	🟥	🟥	🟩	🟥	🟩	🟩	🟩	🟥	10
Rabensteiner (1975)	🟩	🟩	🟥	🟩	🟥	🟥	🟩	🟥	🟥	🟥	🟥	🟥	🟥	🟥	🟩	🟥	🟩	🟥	🟩	🟥	7
Rohovsky (1965)	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟥	🟥	🟩	🟥	🟩	🟥	8
Salihovic et al. (2012)	🟩	🟩	🟥	🟩	🟥	🟥	🟥	🟥	🟩	🟥	🟥	🟥	🟥	🟥	🟩	🟩	🟥	🟥	🟩	🟥	7
Schieve et al. (2009)	🟩	🟩	🟥	🟩	🟩	🟩	🟥	🟩	🟩	🟩	🟩	🟩	🟥	🟥	🟩	🟩	🟩	🟩	🟩	🟥	15
Schlanger and Gottsleben (1957)	🟩	🟩	🟥	🟩	🟩	🟩	🟩	🟩	🟥	🟩	🟥	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟩	🟥	16
Stansfield (1990)	🟩	🟥	🟥	🟩	🟥	🟥	🟥	🟥	🟩	🟩	🟩	🟩	🟥	🟥	🟥	🟩	🟩	🟥	🟩	🟥	9
Total score per item	14	12	0	14	3	3	5	10	9	11	7	6	3	4	11	7	12	6	14	2	

The AXIS critical appraisal tool for cross-sectional studies (Downes et al., 2016) was used to assess study quality. Green indicates an item score of 1 (i.e., a positive evaluation). Red indicates an item score of 0 (i.e., a negative evaluation).

2018). It is therefore likely that the high rate of occurrence is related to an increased vulnerability to stuttering associated with the biomedical condition of Down syndrome and is not a characteristic of Down syndrome.

The occurrence estimates were high (approximately 18–25%) across school-aged and adult participants, while in preschool-aged participants, the occurrence estimate was low relative to the cumulative incidence in typically developing children. These results indicate an opposite pattern to that commonly observed in typically developing individuals, in which the occurrence usually is higher in the preschool years and decreases with age (Bloodstein et al., 2021). Even though the results of the present study may indicate higher occurrences of stuttering in older individuals with Down syndrome, the results do not necessarily mean that the occurrence of stuttering in this population increases with age. As there are no studies following the same participants across time, the results represent the occurrence of stuttering in different age groups and not the developmental pattern of stuttering. The occurrence of stuttering is expected to vary with the sampled age group (Samson, 2022). However, several aspects related to the design of the included studies make it difficult or even impossible to discuss differences in occurrence estimates across age groups. The combination of non-probability sampling techniques, small sample sizes, and age-spread samples do, for example, mean that there are uneven numbers of participants across age groups. Thus, occurrence estimates may suffer from an overrepresentation of age groups where stuttering is more or less common, or findings simply being coincidental as single participants may have a large influence on the results.

A minority of the studies in this review provided information on the occurrence of stuttering by gender. The synthesis of the results of these studies suggests that stuttering was twice as common in male participants as in female participants. However, the gender balance seems to be similar across studies (based on total *n*). This result is in line with findings from studies of the typical adult population, in which more male participants are found to stutter than female participants (gender ratio of between 2:1 and 4:1; Craig et al., 2002). In typically developing young children, the gender distribution is more balanced (Samson, 2022), but more male participants than female participants still stutter (gender ratio of 1.6:1; Sjøstrand, 2022). Whether this asynchrony between genders occurs because of skewed birth figures for boys and girls, because male participants are more vulnerable to stuttering or is related to the indicators of stuttering, is unknown.

Conclusions regarding the characteristics of stuttering are restricted by the specificity of the information provided in the included studies. For example, although studies have consistently reported a high occurrence of stuttering, the current review cannot provide much information regarding the distribution of disfluency types, as the dissertation by Keane (1970) is the only study that provides information about the disfluency types identified in participants who stutter. However, the existence of repetitions, prolongations, and blocks, which are usually included in the evaluation of stuttering in typically developing individuals (Bloodstein et al., 2021), were common identifiers of stuttering across studies. Thus, the results indicate that individuals with Down syndrome display the same speech behaviors as typically developing individuals who stutter.

Furthermore, the focus on the potential adverse effects of stuttering is very limited in the reviewed studies. Even though the participants in the current review exhibited stuttering severity in the mild-to-moderate range, this does not mean that the potential effects of stuttering are not extensive, as studies demonstrated no significant relationship between stuttering frequency and negative feelings about communication (Erickson and Block, 2013).

4.1 Implications for practice and research

As stuttering is common, can cause negative reactions (Jackson et al., 2014; Bray, 2017), and have negative consequences for communication in individuals with Down syndrome (Evans, 1977; Maessen et al., 2022), practitioners must refer those with disfluent speech to speech and language therapists for assessment and, eventually, treatment. Thus, validated assessment procedures and research-based treatments developed especially for this population should be trialed in future research. In addition, information that is relevant to teachers, parents, and healthcare professionals should be developed to inform them about the high occurrences of stuttering in individuals with Down syndrome and when referral for the evaluation of stuttering is appropriate.

4.2 Limitations

We want to highlight four limitations related to the occurrence estimates of the present study. Because the measures and sample characteristics differ across the studies, it is not straightforward to conclude the occurrence of stuttering. The findings should be interpreted as estimates, as they are likely influenced by (1) sample size, (2) how stuttering is operationalized and assessed, (3) the language proficiency of the participants, and (4) at what age stuttering is measured.

Furthermore, because this review is based on only concurrent data regarding stuttering, we do not know how the occurrence rate by age relates to the tractability of stuttering in this population. For example, it is unknown whether those who stutter at young ages continue to stutter later in life. To answer this question, longitudinal studies are needed. Also, as this review only included studies that investigated the occurrence of stuttering, studies with samples preselected based on fluency status have not been included. This implies that there may be more available research investigating the characteristics of stuttering in individuals with Down syndrome that has not been included in this study. Finally, it should be mentioned that some of the confidence intervals in our inter-rater reliability analysis are wide, indicating a limitation in the precision of these estimates. This uncertainty in some of the estimated effect sizes may reflect the low number of studies in this review (Hazra, 2017), as each disagreement has a large influence on the effect size and its confidence level.

5 Conclusion

The results of this systematic review show a high rate of stuttering occurrence in individuals with Down syndrome, independent of assessors, when interpreted against results derived from studies on occurrence estimates in typically developing individuals. This applies to both male and female participants, but the relative proportion of male participants among stutterers is higher. Furthermore, the occurrence in the school-aged and adult participant groups is especially high. While the operationalization of stuttering varied across the studies, the identification of repetitions, prolongations, and blocks was typically included. Stuttering was commonly judged to be mild-to-moderate, and secondary behavior was found when measured.

Author contributions

SH and K-AN: conceptualization, methodology, formal analysis, resources, investigation, data curation, writing–original draft preparation, and writing–review and editing. SH: visualization and project administration: K-AN and SH: funding acquisition. All authors have read and agreed to the published version of the manuscript.

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Conflict of interest

Publisher's note

Supplementary material

The Supplementary material for this article can be found online at: https://www.frontiersin.org/articles/10.3389/fpsyg.2023.1176743/full#supplementary-material

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Note: *Included in review.

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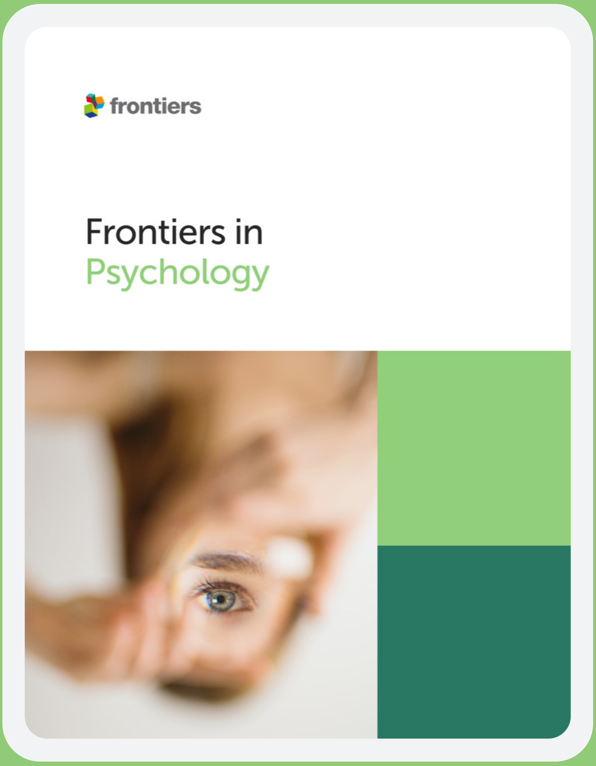
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