

PSYCHOLOGY AND NEUROPSYCHOLOGY OF PERCEPTION, ACTION, AND COGNITION IN EARLY LIFE

EDITED BY: Silvio Ionta, Els Ortibus and Jane Elizabeth Aspell

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PSYCHOLOGY AND NEUROPSYCHOLOGY OF PERCEPTION, ACTION, AND COGNITION IN EARLY LIFE

Topic Editors:

Silvio Ionta, University of Lausanne, Switzerland

Els Ortibus, KU Leuven, Belgium

Jane Elizabeth Aspell, Anglia Ruskin University, United Kingdom

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Editorial: Psychology and Neuropsychology of Perception, Action, and Cognition

Jane E. Aspell¹, Els Ortibus² and Silvio Ionta^{3*}

¹ School of Psychology and Sport Science, Anglia Ruskin University, Cambridge, United Kingdom, ² Locomotor and Neurological Disorders, Department of Development and Regeneration, KU Leuven, Leuven, Belgium, ³ Sensory-Motor Lab (SeMoLa), Department of Ophthalmology-University of Lausanne, Jules Gonin Eye Hospital-Fondation Asile des Aveugles, Lausanne, Switzerland

Keywords: brain, vision, movement, multisensory, sensorimotor

Editorial on the Research Topic

Psychology and Neuropsychology of Perception, Action, and Cognition

Translating multidisciplinary scientific knowledge into unified psycho-educational practices can improve the restoration and establishment of basic functions, such as using tools or interacting with others. For instance, low vision has a tremendous impact on writing, navigating, or playing in groups, but current interventions rely on specialized knowledge from different areas typically operating in isolation.

Following from the symposium “NeuroPedagogy of Vision and Beyond”, held at the Fondation Asile des Aveugles in May 2021, with the support of the Center Pédagogique pour élèves Handicapés de la Vue (CPHV) and the Frontiers Publishing Group, the present Research Topic collects 14 articles (11 original studies, three reviews) by world-leading neuroscientists, pedagogues, neuropsychologists, clinicians, and developmental psychologists. The articles describe state-of-the-art behavioral, psychophysical, and brain imaging studies of sensory-motor-cognitive loops in health and disease, within three main topics: perception, action, cognition (**Table 1**).

Within the “*perception*” axis, Yang et al. provide new evidence about the development of binocular suppression mechanisms. The authors used a continuous flash suppression task to induce a conflict between the visual input (one perceptually dominant and one perceptually non-dominant image) delivered to one and the other eye, in 2–5-month-old infants. Only younger infants showed to perceive the non-dominant image, indicating that about 3 months the binocular suppression mechanisms are not fully formed yet.

Within the “*cognition*” axis, Solovieva and Quintanar highlight the influence of cultural factors on the so-called first year developmental crisis, involving radical psychological changes constituting the basis of the following motor, cognitive, social developments (Solovieva and Quintanar). This paper provides important information for optimizing child-adult interactions while building a reliable psychological context for the child’s subsequent development. In the same “*cognitive*” vein, Ling et al. show that the properties of the task used to assess conditional reasoning skills can importantly affect the performance and, therefore, the establishment of the minimum age for appropriate conditional reasoning. The authors used a modified version of the dimensional change card sort (DCCS), including color as a key feature of target objects. While the DCCS task is typically solved around the age of 5, this manipulation enabled 3-year-old children to succeed, highlighting the importance of accounting for possible implicit biases of commonly accepted procedures (Ling et al.). Using a similar experimental approach, Zacharov et al. illustrated the impact of autism on cognitive flexibility. The authors administered a DCCS task, in combination with a reverse categorization task and a non-verbal cognitive age assessment, in children with and without autism. Autism was associated with worse performance in the DCCS task and disrupted

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Edited and reviewed by:

Lutz Jäncke,
University of Zurich, Switzerland

*Correspondence:

Silvio Ionta
ionta.silvio@gmail.com

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TABLE 1 | Classification of included articles as a function of the RT's topics (P, Perception; C, Cognition; A, Action).

#	Type	1st Author	Frontiers in	Class	Technique	Age
1	Exp	Yang	Psychology	P	Behaviour	2–5 months
2	Review	Solovieva	Psychology	C	Behaviour	1 year
3	Exp	Ling	Psychology	C	Behaviour	3 years
4	Exp	Zacharov	Psychology	C	Behaviour	3–6 years
5	Exp	Alghamdi	HumNeur	P/C	Behaviour	5–7 years
6	Exp	Ye	Psychology	A/C	Behaviour	1–3 years
7	Exp	Farran	HumNeur	A/C	Behaviour	5–11 years
8	Exp	Fitamen	Psychology	A/C	Behaviour	5 years
9	Exp	Weibley	BehavNeur	A/C	fNIRS, behaviour, cognitive	8–14 months
10	Exp	Guan	Psychology	A/P	EEG, behavior	9–10 years; adults
11	Exp	Esposito	HumNeur	A/P	Behaviour	adults
12	Exp	Micheletti	HumNeur	A/P	Behaviour	5–12 years
13	Review	Ionta	HumNeur	A/P	Brain, behaviour	0–65 years
14	Review	Farah	Psychology	A/P/C	Brain, behaviour	0–65 years

the correlation between mental age and performance in both tasks. However, when the two groups were matched by mental age, their performance in the two tasks was not significantly different, highlighting the importance of fine classifications and precise evaluations of the methods used to assess cognitive skills.

Within the “perception-cognition” axis, Alghamdi et al. investigated the relationship between global intelligence and visual processing by measuring the speed of visual inspection, visuo-verbal interactions, and visuomotor control in 5–7-year-old children. The latter two were found to correlate with non-verbal intelligence scores and years of education and as such, might be possible targets in educational programs.

Along the line of Zacharov et al., but within the “action-cognition” axis, a specific focus on autism was adopted also by Ye et al. to illustrate the characteristics of gesture production in 2–4 year-old children with autism. Children with autism showed less behavioral regulation, social interaction, and joint attention gestures. However, similar to Zacharov et al., correcting the gesture performance by the communication score changed the outcomes. Without correction, children with autism exhibited fewer gestures both with and without accompanying vocalization. With the correction, only the production of gesture without vocalization was lower in children with autism with respect to controls. In line with Ling et al. and Zacharov et al., the work by Ye et al. also underlines the importance of finely evaluating the assessment tools themselves, and not only the assessed populations. The relationship between cognitive and motor skills permeates three other articles. First, Farran et al. investigated the impact of physical disability on spatial cognition. They administered mental rotation, spatial programming, and virtual navigation to two groups of physically impaired children (differentiated by the need or not of wheelchair use) and controls. The performance of the two groups with physical disabilities was lower than controls, but did not vary between wheelchair users or not. This suggests that physical disability affects spatial cognition to a degree large enough that further differentiations as

function of contextual factors (wheelchair) are minimal. Second, Fitamen and Camos show the benefits of motor activity on subsequent memory processes. These authors asked 5-year-old children to perform a memory task after a game-like and an exercise-like motor activity (in counterbalanced order) which both involved the objects used in the subsequent memory task. Children performed better in the exercise-like memory trials when they performed the exercise-like motor activity before the game like activity. Conversely, when they had the game-like motor activity before the exercise-like motor activity, there was no difference in their performance with exercise-like and game-like memory trials. This shows the superiority of game-like activities in establishing better-lasting memories. Third, focusing on the same age range and complementing behavioral observations of Solovieva and Quintanar, Weibley et al. described the cortical correlates of motor and cognitive skills. The authors monitored brain activity in the prefrontal cortex of children aged between 8 and 14 months while they were performing active and passive motor and attentional tasks. Within each category, active tasks were associated with higher prefrontal activity compared to passive tasks, highlighting the importance of active involvement in daily activities for motor-cognitive development (Weibley et al.).

Within the “action-perception” axis, Guan et al. studied the effects of motor activity on a subsequent perceptual task in 9–10-year-old children and adults (Guan et al.). Using EEG, they investigated the influence of a previous handwriting condition on the neural correlates of a subsequent visual word recognition task. They showed that in adults, but not in children, the brain activity during the visual perception task was lateralized toward the hemisphere dominant for language (left). The authors discuss their findings with respect to the importance of maintaining handwriting training in the digital era. A similar focus was adopted by the study by Esposito et al. that analyzed the consequences of early visual deprivation on the development of head-trunk coordination movements. The authors recorded

movement kinematics from the head and trunk of young adults who were congenitally blind and controls performing a head-pointing task while voluntarily immobilizing (or moving) the trunk. While movement analysis showed a head-trunk coordination impairment in congenitally blind participants, their performance in the task was not significantly different from controls. This supports the plasticity of visuo-motor interactions, in that compensatory mechanisms can enable the achievement of goals through alternative strategies. In the same visuo-motor vein, Micheletti et al. examined the relationship between motor impairment and visual skills. In over 100 children with developmental coordination disorder (DCD) they compared the sensitivity to global motion and global static form stimuli to those of controls. Results showed that the performance of children with DCD in the global motion task was worse than controls, and that motor impairment was linearly correlated with global form sensitivity and presented a quadratic correlation with global motion sensitivity. The authors discuss these findings with reference to the differentiation between dorsal (motion) and ventral (form) stream functions, which seem differentially affected by motor impairment. Such a neuro-behavioral approach in the context of visuo-motor interactions is extended by two review papers. The first one summarizes over 300 papers about neuropsychological evidence on (i) the neural correlates of vision, (ii) anatomo-functional brain dynamics associated with the development of visual, motor, and visuo-motor skills in health and disease and across the life span (Ionta), and (iii) visuo-motor perspectives on relatively lower-level and more complex syndromes, such as strabismus, akinetopsia, DCD, and hemispatial neglect. The second one establishes a “*perception-action-cognition*” bridge, focusing on the neural and behavioral

peculiarities of executive function in dyslexia (Farah et al.). Summarizing evidence from about 200 papers, this review highlights the importance of examining executive functions as possible early predictors of following reading/speaking deficits.

Overall, by strengthening the understanding of the neural bases of developmental disorders, the insights derived from the present Research Topic will hopefully provide a solid background to support interdisciplinary discussions among experts in sensory and/or motor disorders in the context of neuro-behavioral rehabilitation and training.

AUTHOR CONTRIBUTIONS

All authors listed have made a substantial, direct, and intellectual contribution to the work and approved it for publication.

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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The Development of Binocular Suppression in Infants

Jiale Yang^{1*}, So Kanazawa² and Masami K. Yamaguchi³

¹ Research and Development Initiative, Chuo University, Tokyo, Japan, ² Department of Psychology, Japan Women's University, Kanagawa, Japan, ³ Department of Psychology, Chuo University, Tokyo, Japan

Little is known about the time of development of binocular suppression. In the present study, we evaluated the emergence of binocular suppression in infants by using continuous flash suppression (CFS, Tsuchiya and Koch, 2005). In our experiment, one eye of infants was presented with a static face image at one side of the screen, while another eye was presented with dynamic Mondrian patterns in full screen. Adult observers confirmed that the static face image was consciously repressed by the changing Mondrian patterns. If binocular suppression was functional, the infants would not perceive the face and thus would not show any preference in the experiment. However, if binocular suppression in the infants was not yet acquired, they would perceive the face and the Mondrian patterns at the same time and would thus show preference for the side where the face was presented. The results showed that infants aged 2–3 months, but not those aged 4–5 months, detected the position of the face. Furthermore, this detection was not due to weak contrast sensitivity to the dynamic Mondrian mask. These results indicated that the immature binocular visual system may perceive different images from different eyes simultaneously and that infants may lose this ability after establishing binocular suppression at 4–5 months of age.

Keywords: binocular suppression, infant, preferential looking paradigm, visual development, continuous flash suppression

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Jane Elizabeth Aspell,
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United Kingdom

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Sarah Jessen,
University of Lübeck, Germany
Teresa Mitchell,
Brandeis University, United States

*Correspondence:

Jiale Yang
jjale.yang.ac@gmail.com

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INTRODUCTION

Several studies on binocular vision in infants have found that most infants have an average stereopsis onset between the ages of 2 and 4 months (Fox et al., 1980; Held et al., 1980; Petrig et al., 1981). Developmental stereopsis has been investigated by measuring the emergence of the sensitivity to binocular disparity (Held et al., 1980; Petrig et al., 1981; Skarf et al., 1993; Birch and Petrig, 1996; Kavšek, 2013a). In these studies, two paradigms have been applied, namely, the measurement of visual evoked potentials (VEPs) and the measurement of looking time to the stimuli containing or without horizontal disparity information. These studies suggested that sensitivity to horizontal disparity emerges after 3 months of age.

Binocular rivalry, another perceptual phenomenon of binocular vision, has been investigated by testing whether the infant can discriminate between fusible and rivalrous stimuli (Birch et al., 1985; Shimojo et al., 1986; Gwiazda et al., 1989; Thorn et al., 1994; Brown and Miracle, 2003; Kavšek, 2013b). In this method, two stimuli were presented dichoptically to infants. One stimulus consisted of an interocularly identical pattern (fusible stimulus) and the other of an interocularly different pattern (rivalrous stimulus). The forced-choice preferential looking (FPL) method (e.g., Teller, 1979) was used to examine whether the infants showed a preference for certain stimuli, which is

regarded as discrimination between the fusible and rivalrous stimuli. These studies reveal that the infants looked longer at the fusional stimuli than the rivalrous ones after an average age of 2 months, suggesting that binocular rivalry emerges at a similar period as stereopsis.

These previous studies using preferential looking methods to test the discrimination between fusible and rivalrous patterns depend on whether infants have a spontaneous preference for the fusible pattern or rivalry pattern. If infants show a preference for certain stimuli, this indicates that the infants can detect fusible stimuli from rivalrous stimuli. However, a null result in the younger group cannot be interpreted as that these infants do not perceive the binocular rivalry, because it is possible that these infants would show no spontaneous preference for the fusible pattern or rivalrous pattern. Therefore, the emergence of preference shown by previous studies could not be considered as the developmental onset of the binocular rivalry.

In the present study, we evaluated the development of binocular rivalry in infants by using continuous flash suppression (CFS, Tsuchiya and Koch, 2005), which does not depend on the spontaneous preference for either the fusible or the rivalrous pattern. Hence, it can avoid a null result as commonly observed in prior studies, which is hard to interpret with respect to whether infants do show binocular rivalry or not. In the CFS procedure, a target stimulus is continuously presented to one eye, while continuous flashing of random Mondrian images is presented to the other eye. This CFS prevents participants from seeing the target image. Different from classical binocular rivalry, the target can be completely suppressed for over 1 min by using the CFS paradigm (Tsuchiya and Koch, 2005). Thanks to this long suppression time, CFS allows researchers to manipulate conscious perception. Therefore, CFS is used as a powerful tool in the aspect of consciousness studies (for a review, see Axelrod et al., 2015). For instance, a recent adult study showed that images of dominant and untrustworthy faces, compared to neutral faces, took a longer time to emerge to awareness, suggesting that information about personality characteristics can be processed outside of awareness (Stewart et al., 2012). If we could demonstrate that CFS can be utilized in infants successfully, this would not only give us an opportunity to investigate binocular suppression in infants but also indicate that CFS can be used in consciousness studies in infants.

In the present study, one eye of each infant was presented with a static face image at one side of the screen, while the other eye was presented with dynamic Mondrian patterns over the entire screen (**Figure 1**). If binocular suppression has developed, the infants would not perceive the face similarly as adults and thus would show no face preference in the experiment. If the infants had not acquired binocular suppression yet, they would perceive the face and the Mondrian patterns simultaneously and would thus show preference for the face. We used the face as a target because a visible face elicits reliable attentional biases toward it even in newborns (Johnson et al., 1991; Mondloch et al., 1999; Cassia et al., 2004; Di Giorgio et al., 2012). If the face stimulus was visible for infants in present study, the infants would perceive a face mixed with dynamic Mondrian patterns. We hypothesized that this “mixed face” would attract the infants’ attention and

give rise to preferential looking toward the side where the “mixed face” was located.

EXPERIMENT 1

In the first experiment, we investigated the development of binocular suppression in infants by using CFS.

MATERIALS AND METHODS

Participants

Fifteen infants aged 2–3 months (7 male, 8 female, mean age = 73.6 days, and age range 51–98 days) and 15 infants aged 4–5 months (6 male, 9 female, mean age = 137.8 days, and age range 105–161 days) were included in the study. Although 25 other infants were tested in Experiment 1, they were excluded from the analysis because of fussiness ($n = 7$), side bias of more than 90% ($n = 16$), or technical problems ($n = 2$). All infants were recruited through advertisements in the newspaper and were full-term at birth and healthy at the time of the experiment. Ethical approval for this study was obtained from the ethical committee at Chuo University. Moreover, the experiments were conducted according to the principles of the Helsinki declaration. Written informed consent was obtained from the parents of the infants prior to the start of the experiment.

Apparatus

During the experiment, each infant sat on his or her parent’s lap in the experimental booth. A 22-inch three-dimensional liquid-crystal display (3D-LCD) monitor (ZM-M220W; Zalman Tech Co Ltd.) that displayed all the stimuli was placed in front of the infant, at a distance of about 40 cm. Infants wore circular 3D glasses to watch the stimuli during the experiment. The center of the monitor was at the infant’s eye level, and its resolution was set at $1,680 \times 1,050$ pixels. The infant’s looking behavior was recorded through a video camera set under the monitor. Behind the experimental booth, the infant’s behavior was also observed via a TV monitor.

Stimuli

Two different images were dichoptically presented to both eyes of the infants (**Figure 1**). One eye was presented with a neutral grayscale face image, which was generated by averaging different 20 Asian women’s faces, on a gray background, while the other eye was presented with a series of color dynamic Mondrian patterns refurbished at 10 Hz in full screen. The face image subtended 10.2×6.3 degrees and was randomly situated on either the left or the right side of the screen. The mean luminances of the face image, the Mondrian patterns, and the background were 17.6, 39.6, and 12.1 cd/m^2 , respectively. The stimuli were presented for 3 s in each trial. Two adult observers have confirmed that the face presented to one eye can be completely suppressed by dynamic Mondrian patterns presented to the other eye during CFS.

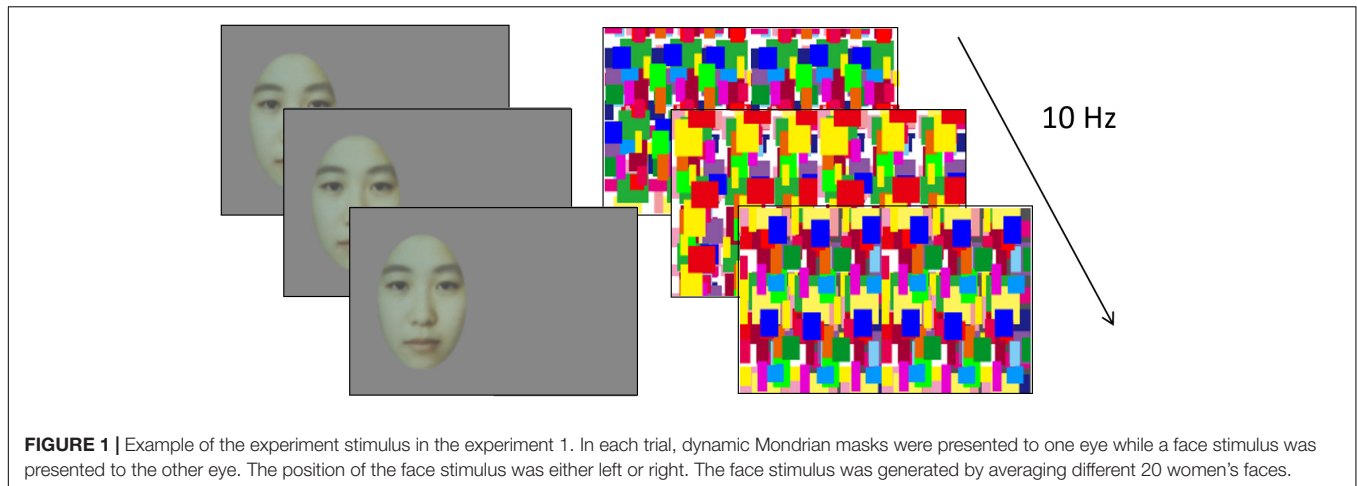


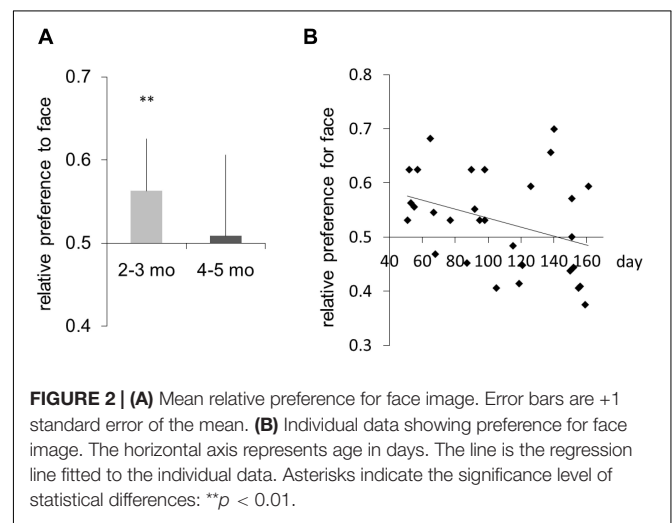
FIGURE 1 | Example of the experiment stimulus in the experiment 1. In each trial, dynamic Mondrian masks were presented to one eye while a face stimulus was presented to the other eye. The position of the face stimulus was either left or right. The face stimulus was generated by averaging different 20 women's faces.

Procedure

The FPL paradigm consisting of 32 trials was used in our experiment. A fixation figure was shown in the center of the monitor accompanied by a short beep sound prior to each trial to attract the infant's attention. After confirming that the infant was looking at the fixation figure, the experimenter started the trial. In each trial, the stimulus was presented for 3 s. The position of the face image was randomly assigned in each trial. The parents were instructed to close their eyes during the experiment. An observer, who did not know the stimulus identity, judged whether the infant looked at the left half or at the right half of the screen based on an offline video movie. When only "no-looking" was recorded, the trial was excluded. Forty percent of the trials were recorded by a second trained observer. The interrater reliability of the two observers was calculated by intraclass correlation coefficient (ICC) using SPSS statistical package version 23 (ICC = 0.90 with 95% confidence interval = 0.86–0.93).

Results

The mean number of completed trials per participant was 28.58 (SD = 5.58). Preference scores were calculated as the probability of correct judgment for the position of the face image. We regarded these preferences as the detection of the face image. **Figure 2A** shows the average preference scores for the face image (2- to 3-month-old infants: mean = 0.569, SD = 0.05; 4- to 5-month-old infants: mean = 0.508, SD = 0.09). Two-tailed one-sample *t*-tests against a chance level of 0.5 were conducted for each age group. Significant preference for the face image was observed in the 2- to 3-month-old infants [one-sample *t*-test (vs. chance level, 0.5), $t(14) = 3.87$, $p < 0.01$, $d = 0.99$; a *post hoc* power analysis showed that the study had above 94% power to detect a significant difference at $p < 0.05$] but not 4- to 5-month-old or 5- to 6-month-old infants [$t(14) = 0.33$, n.s.]. An independent *t*-test revealed that the preference scores were different in the two age groups [$t(28) = 2.11$, $p < 0.05$, $d = 0.77$; a *post hoc* power analysis showed that the study had above 80.8% power to detect a significant difference at $p < 0.05$]. Moreover, there was a strong negative correlation between age and preference score ($r = -0.349$,



$p < 0.05$; **Figure 2B**). These results suggest that 2- to 3-month-old infants are sensitive to masked face images that adults cannot perceive. The immature binocular visual system may probably perceive different images from different eyes simultaneously, and the infant may lose this ability after establishing binocular suppression at 4–5 months of age.

It is well known that spatial and temporal contrast sensitivity is significantly lower in infants compared with adults (Teller, 1998). It is possible that the 2- to 3-month-old infants might have been able to detect the face image due to their low sensitivity to the dynamic change in the mask in Experiment 1. Therefore, the dynamic Mondrian patterns in the present study may not have enough intensity to generate interocular suppression. Hence, we tested this possibility in Experiment 2.

EXPERIMENT 2

In Experiment 2, we examined whether the 2- to 3-month-old infants have enough contrast sensitivity to perceive the dynamic Mondrian patterns. A gray background was presented to one eye,

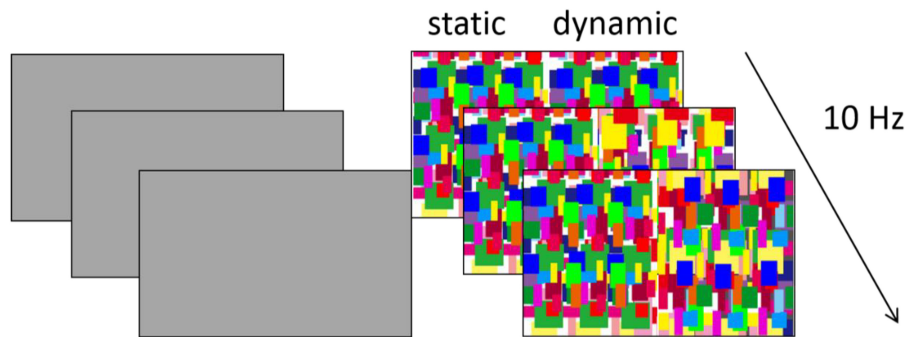


FIGURE 3 | Example of the experiment stimulus in Experiment 2. In each trial, dynamic and static Mondrian masks were presented to one eye, while a gray background was presented to the other eye. The position of the static/dynamic stimulus was randomized.

while static Mondrian patterns and dynamic Mondrian patterns were presented to the other eye side by side and simultaneously. If the infants had enough contrast sensitivity to perceive the dynamic Mondrian patterns, they would detect the change and show a preference for the side of dynamic Mondrian patterns.

Participants

Ten infants aged 2–3 months (7 male, 8 female, mean age = 78.9 days, and age range 54–89 days) participated in the study. Although eight other infants were tested in Experiment 2, they were excluded from the analysis because of fussiness ($n = 6$) or side bias of more than 90% ($n = 2$).

Stimuli

Two different images were shown dichoptically to both eyes of the infants. One eye was presented with a gray background with a luminance of 17.6 cd/m^2 . The other eye was presented with a half side of dynamic Mondrian patterns, which was identical to that from Experiment 1, and a half side of static Mondrian patterns, which represented one frame of the dynamic Mondrian patterns (Figure 3). The dynamic Mondrian patterns alternated at 10 Hz, while stimuli were presented for 3 s in each trial.

Apparatus and Procedure

The apparatus and procedure were identical to those used in Experiment 1. Each infant was presented with 32 trials in which the position of the dynamic Mondrian patterns was randomized. Forty percent of the trials were recorded by a second trained observer. The interrater reliability of the two observers was calculated by ICC using SPSS statistical package version 23 (ICC = 0.91 with 95% confidence interval = 0.87–0.95).

RESULTS

The mean number of completed trials per participant was 28.90 (SD = 5.13). Preference for the dynamic Mondrian patterns was observed in the 2- to 3-month-old infants (mean = 0.508, SD = 0.09; Figure 4). A one-sample t -test showed that the infants significantly preferred the dynamic Mondrian patterns over chance level [$t(9) = 4.70, p < .01, d = 1.57$; a *post hoc* power

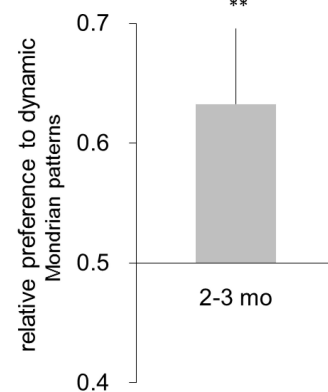


FIGURE 4 | Mean relative preference for the face image. Error bars are +1 standard error of the mean. Asterisks indicate the significance level of statistical differences: $**p < 0.01$.

analysis showed that the study had above 99% power to detect a significant difference at $p < 0.05$). These results suggested that 2- to 3-month-old infants could detect the change in the dynamic Mondrian patterns. Therefore, it is unlikely that the detection of the face image by the 2- to 3-month-old infants in Experiment 1 was simply the consequence of their poor spatiotemporal contrast sensitivity.

GENERAL DISCUSSION

The present study investigated the development of binocular suppression among 2- to 5-month-old infants by using the CFS technique. In Experiment 1, we investigated whether infants could perceive a face image masked by dynamic Mondrian patterns. If the function of binocular suppression has not emerged yet, infants should detect the face image and show a significant preference for side where the face image was presented. The results revealed that only the 2- to 3-month-old infants showed a preference for the face side. In Experiment 2, we confirmed that the 2- to 3-month-old infants had enough sensitivity to perceive the change

in the dynamic Mondrian patterns. This has confirmed that their ability to detect the masked face image did not stem from weak contrast sensitivity. These results indicated that the immature binocular visual system in 2- to 3-month-old infants may allow them to perceive different images from different eyes simultaneously, while adults' visual perception would be completely suppressed by the input from the other eye.

The infant may lose the ability to perceive different images from different eyes simultaneously after establishing binocular suppression at 4–5 months of age. Declines in developmental trajectories have been found in many aspects of perception (for a review, see Lewkowicz and Ghazanfar, 2009). For instance, a recent study has reported that 3- to 4-month-old infants react directly to low-level image features that adults might ignore and that this ability would be lost after 5 months of age (Yang et al., 2015). Although some functions show regressive developmental processes, the shifts of the computational scheme reflect the development in the visual system of young infants.

Using CFS, we found different results from those studies (i.e., Brown and Miracle, 2003; Kavšek, 2013b) using preferential looking methods to test the discrimination between fusible and rivalrous patterns. Infants aged 2–3 months old showed a reversal preference for the rivalrous patterns in our study; in contrast, infants aged 2–4 months old preferred the fusible patterns rather than the rivalrous patterns in Brown and Miracle (2003) and Kavšek (2013b). It must be noted that the stimuli were completely different in these studies: both sides of the stimulus were rivalrous patterns in present study, but those in previous studies were fusible patterns vs. rivalrous patterns. Even though we knew that 2- to 3-month-old infants might prefer the fusible pattern from previous studies, it is difficult to predict how infants perceive the rivalrous patterns. It is possible that these infants perceive only one image from one eye at a time like adults. Another possibility is that these infants' perceived image is an unstable mixture of the two images from two eyes, analogous to the transition state of binocular rivalry in adults, because the immature binocular functioning might not have been sufficient for providing the energy to suppress the inputs from other eye completely. Our results suggest that the latter is more plausible because these infants could detect the face during CFS, indicating that the dynamic Mondrian pattern can only suppress a part of the inputs from the other eye. Therefore, the development of binocular rivalry seems to be a continuous process after birth. The 2- to 3-month-old infants might experience an incomplete form of binocular rivalry, perceiving an unstable mixture from two eyes, and then develop an adult-like binocular rivalry after 3 months of age.

Recently, it has been reported that individuals with autism spectrum disorder (ASD) demonstrated a slower rate of binocular rivalry alternations with longer durations of mixed percepts that matched typically developing infants, which might be caused by the lack of balance between cortical excitation and inhibition (Robertson et al., 2013). The imbalance between

cortical excitation and inhibition in young infants may impair interocular suppression, which permits them to perceive the face image under a dynamic Mondrian pattern. Promising future research would be to explore whether newborns who would later be diagnosed with ASD would have a different binocular rivalry or suppression.

In the present study, we found that 2- to 3-month-old infants could perceive the face target during CFS. In addition to the possibility that the immature binocular visual system allows 2- to 3-month-old infants to perceive different images from different eyes simultaneously, subcortical processing could be involved in face detection under binocular suppression in 2- to 3-month-old infants. Previous studies show that newborns can detect faces while the visual cortex is still immature, indicating that subcortical pathways are involved in face detection in infants less than 3 months of age (Johnson et al., 1991; Mondloch et al., 1999; Cassia et al., 2004; Di Giorgio et al., 2012; for a review, see Johnson, 2005). Furthermore, a recent study demonstrated that subcortical face processing affects face detection in 2-month-old infants (Nakano and Nakatani, 2014). On the other hand, functional magnetic resonance imaging in adults revealed that the subcortical region responds to invisible face stimuli under CFS (Jiang and He, 2006; Troiani and Schultz, 2013). Taken together, these pieces of evidence indicate a possibility that the face detection of 2- to 3-month-old infants reflects subcortical processing of the face.

To conclude, the current study provided the first investigation of binocular suppression in infants using CFS. Our findings suggested that infants aged 2–3 months could detect the target under CFS and that by 4 months of age, binocular suppression emerges, resulting consequently in the inability of 4- to 5-month-old infants to perceive the target during CFS.

DATA AVAILABILITY STATEMENT

The datasets presented in this study are available from the corresponding author, JY, upon reasonable request.

ETHICS STATEMENT

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

AUTHOR CONTRIBUTIONS

JY developed the study concept. All authors contributed to the study design. JY performed testing and data collection, data analysis and interpretation under the supervision of SK and MY, and drafted the manuscript. SK and MY

provided critical revisions. All authors approved the final version of the manuscript for submission.

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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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Children Only 3 Years Old Can Succeed at Conditional “If, Then” Reasoning, Much Earlier Than Anyone Had Thought Possible

*Daphne S. Ling, Cole D. Wong and Adele Diamond**

Developmental Cognitive Neuroscience Program, Department of Psychiatry, The University of British Columbia, Vancouver, BC, Canada

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Gian Marco Marzocchi,
University of Milano-Bicocca, Italy

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*Correspondence:

Adele Diamond
adele.diamond@ubc.ca

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That conditional, if-then reasoning does not emerge until 4–5 years has long been accepted. Here we show that children barely 3 years old can do conditional reasoning. All that was needed was a superficial change to the stimuli: When color was a property of the shapes (line drawings of a star and truck) rather than of the background (as in all past conditional discrimination [CD] testing), 3-year-olds could succeed. Three-year-olds do not seem to use color to inform them which shape is correct unless color is a property of the shapes themselves. While CD requires integrating color and shape information, the dimensional change card sort (DCCS) task requires keeping those dimension cognitively separate – inhibiting attention to one (e.g., shape) when sorting by the other (e.g., color). For DCCS, a superficial change to the stimuli that is the inverse of what helps on CD enables 3-year-olds to succeed when normally they do not until $\sim 4\frac{1}{2}$ years. As we and others have previously shown, 3-year-olds can succeed at DCCS when color is a property of the background (e.g., a white truck on a red background), instead of a property of the stimulus (e.g., a red truck on a white background, as in standard DCCS). Our findings on CD and DCCS suggest that scaffolding preschoolers’ emerging conceptual skills by changing the way stimuli look (perceptual bootstrapping) enables 3-year-olds to demonstrate reasoning abilities long thought beyond their grasp. Evidently, children of 3 years have difficulty mentally separating dimensions (e.g., color and shape) of the same object and difficulty mentally integrating dimensions not part of the same object. Our present CD findings plus our earlier DCCS findings provide strong evidence against prominent cognitive complexity, conditional reasoning, and graded memory theories for why 3-year-olds fail these two tasks. The ways we have traditionally queried children may have obscured the budding reasoning competencies present at 3 years of age.

Keywords: pull, preschoolers, young children, conceptual understanding, conditional associative learning, dimensional change card sort

INTRODUCTION

In conditional discrimination (CD) tasks with children (Gollin and Liss, 1962; Gollin, 1965; Andrews et al., 2012), which response is correct is conditional on which of two colors is present: Shape A is correct when Color 1 is present and Shape B is correct when Color 2 is present. Psychologists have assumed for over 50 years that the ability to do conditional, if-then reasoning does not develop until roughly 4 to 5 years of age, since children younger than that have consistently failed CD (Gollin and Liss, 1962; Gollin, 1965; Halford et al., 1998a; Halford et al., 2010; Andrews et al., 2012). We show here, however, that children barely 3 years old can succeed at CD (that is, are capable of conditional, if-then reasoning) when superficial stimulus properties are modified. To succeed at a CD task, a child must integrate the two dimensions (color and shape), yet psychologists have traditionally presented CD tasks as if the two dimensions were completely separate. Instead of making color a property of the background or of the outside border on stimulus cards (as in all previous CD experiments with children), we made color a property of the stimuli themselves (the shapes were either Color 1 or Color 2). This manipulation allowed children of 3 years to succeed.

Card sorting (as in the Dimension Change Card Sort [DCCS] task), on the other hand, requires attending only to color information when that dimension is relevant (ignoring shape) or attending only to shape information when that dimension is relevant (ignoring color) and being able to switch from doing one to the other. Correct sorting thus requires a child to separate the same two dimensions that CD requires a child to integrate. Depending on which dimension (shape or color) is currently relevant for sorting the cards, children are supposed to ignore the other. Until we (Diamond et al., 2005) and Kloo and Perner (2005) came along, psychologists had always presented card sorting tasks to children with both color and shape as properties of each stimulus object (e.g., a blue star or a red truck drawn on a stimulus card). Based on the repeated failure of children younger than $4\frac{1}{2}$ to 5 years on the DCCS task, many had concluded that children younger than $4\frac{1}{2}$ years are not capable of conditional reasoning or grasping a hierarchical rule structure (e.g., Frye et al., 1996; Zelazo et al., 2003; Andrews et al., 2012). We hypothesized that if color was a property of the background instead of a property of the stimulus as in canonical (or standard) DCCS testing that children would be able to successfully switch sorting dimensions at a younger age, and indeed that is what we found (Diamond et al., 2005). That simple manipulation enabled children to succeed on the DCCS test at 3 years – 12–18 months earlier than previously reported. Thus separating color and shape in the visual display aided 3-year-olds in conceptually ignoring one dimension when the task required that they focus on the other.

Both CD and DCCS require if, then conditional reasoning and they are both tasks that in their canonical forms 3-year-olds fail but children of 4–5 years pass. They are quite different tasks, however. For example, the rule structure for CD is that when Color 1 is present, Shape A is correct, and when Color 2 is present, Shape B is correct. The rule structure for DCCS is that when the

sorting dimension is color, sort the stimulus card into the bin displaying the same color (ignoring that the shape on the stimulus card and target card over the bin do not match), but when the sorting dimension is shape, sort the stimulus card into the bin displaying the same shape (ignoring that the color on the stimulus card and on the target card do not match). Children are taught the rules for DCCS but not for CD. For CD, children must deduce the rules based on feedback. Feedback is provided on each CD trial but not on any DCCS trial.

We are not hypothesizing that these two tasks require all the same abilities or are in any way isomorphic. We are simply hypothesizing that on these tasks 3-year-olds can be strongly influenced by, and can be heavily dependent on, superficial, surface perceptual features of the stimuli. They can be helped to succeed by changing how things look.

Previously we asked ourselves, “Given the requirements of DCCS what surface modifications to the stimuli might help 3-year-olds?” It seemed to us that since children need to ignore one stimulus dimension when focusing on the other, it would be easier to do that if the two dimensions were not part of the same object (e.g., a drawing of a truck). Also, to the extent that for 3-year-olds a truck is either a truck or it is a red thing, but it cannot be both (Flavell et al., 1986; Perner and Lang, 2002; Kloo and Perner, 2003) separating the two dimensions so color is not an attribute of the truck should be helpful. It turned out we were correct; separating the dimensions did make the task easier for children of 3 and $3\frac{1}{2}$ years (Diamond et al., 2005; Kloo and Perner, 2005).

Similarly, for the present study we asked ourselves, “Given the requirements of CD what surface modifications to the stimuli might help 3-year-olds?” The task requirements are different here than for DCCS. What is needed here is integrating the two dimensions of color and shape. What might help that? We reasoned that integrating them in the stimulus objects themselves should help. Any environment contains lots of perceptual information; how do children know what is relevant and what is not and what to attend to? Having color as an attribute of the truck and star drawings (integrated dimensions) should, we hypothesized, help children realize that color is relevant to the task. Since changing whether color and shape were integrated or separated improved the performance of 3-year-olds on DCCS to roughly the level of 4-year-olds, we hypothesized that changing whether color and shape were integrated or separated would improve the performance of 3-year-olds on CD so that it would roughly approximate the level of 4-year-olds.

Thus, our hypothesis here is that integrating color and shape in the visual display when the task requires conceptually integrating those dimensions (as does CD) should enable 3-year-olds to succeed because it bootstraps the children perceptually in their task of conceptually relating the two dimensions to one another. We tested this and present here the first demonstration that the age of first success on CD can be reduced from 4 or 5 years to 3 years by a surface modification of the stimuli. When color is a property of the stimulus object (i.e., color and shape are integrated as properties of the same object), instead of color appearing as part of the background (separated dimensions), 3-year-olds can succeed at CD.

We broke down our hypothesis into a set of predictions. For all predictions, the block of interest is Block 3. The reason for that is that Blocks 1 and 2 do not require conditioning reasoning. On Block 1 of our CD task, all cards contain blue and the reward is always hidden under the card with a truck drawing (the side of cards being pseudo-randomly varied across trials in all blocks). For Block 2, the reward is always hidden under the card with a drawing of a star, and all the cards contain red. On Block 3, cards containing blue and cards containing red are randomly intermixed over trials, though on each trial both cards contain red or both contained blue. The truck is correct when both cards contain blue (as in Block 1) and the star is correct when both cards contain red (as in Block 2).

Our predictions were:

- (1) First and foremost, children of 3 years would succeed on CD (i.e., succeed on Block 3) when the dimensions of color and shape were integrated in the stimuli. Since we predicted that children of 3 years would perform roughly as well on CD (i.e., roughly as well on Block 3) with integrated dimensions as children of 4 years perform on CD with separated dimensions, we defined success on CD as roughly comparable Block 3 performance by 3-year-olds on integrated dimensions as 4-year-olds show on separated dimensions.
- (2) Children of 3 years would perform significantly better on CD (i.e., on Block 3) when color and shape were integrated in the stimuli than when they were separated.
- (3) We would replicate previous findings (e.g., Gollin and Liss, 1962; Gollin, 1965) that:
 - (a) Children of 3 years will fail CD (i.e., fail to reach criterion in Block 3) when color and shape are separated on the stimulus cards (separated dimensions).
 - (b) Children of 4 years will succeed on CD (i.e., succeed on Block 3) with that same condition (separated dimensions), i.e., they will perform roughly comparably on our CD task with separated dimensions to how other labs (Gollin and Liss, 1962; Gollin, 1965) have found 4-year-olds to perform on CD with separated dimensions when they tested that.
 - (c) In all conditions and at both ages children would succeed on Blocks 1 and 2.

MATERIALS AND METHODS

Participants

A total of 42 children were tested. All children could understand and converse in English and had normal or normal-with-correction hearing and sight. None were taking any medication that affected cognition. None had suffered a concussion or lost consciousness from a fall or blunt trauma to the head. This study was approved by the UBC Behavioral Research Ethics Board (REB# H04-80913), Vancouver Coastal Health Research Institute (V12-80913), and the Vancouver School Board. A parent or guardian of each child gave written informed consent for the child's participation.

Participants were recruited from two age groups: children almost or just barely 3 years old and children almost or just barely 4 years old. They were tested in a StrongStart Centre in the greater Vancouver area (25 children) or in our lab at UBC (17 children). All children were accompanied by a parent, grandparent, or caregiver. The adult chaperone sat behind the child during testing or watched through the lab's one-way mirror. A random subset of sessions was videotaped with permission from the parent or caregiver.

Six children (5 girls and 1 boy) were excluded from data analyses because they appeared unable to grasp how the task worked (that they were to retrieve rewards) or were not interested in it. Five of these children were 3 years old (3 tested on integrated, 2 on separated dimensions [including the one boy]). The sixth child was a 4-year-old girl tested on separated dimensions.

Our data set thus consists of 36 children; 22% were Caucasian, 22% East Asian, 8% Hispanic, 6% South Asian, 11% Mixed Ethnicity, and 6% were other, and 25% did not report their ethnicity. Most children (75%) came from a home where the primary caregiver has a college degree.

In the 3-year-old age group, there were 24 children (44% female). Half were tested on CD with integrated dimensions and half with separated dimensions. The mean age for the 3-year-olds was 3.1 years (SD = 0.16 years; range = 33.5–39.5 months). See **Table 1**.

In the 4-year-old age group, there were 12 children (33% female). They were tested on separated dimensions to see if, when we used the same procedure as have previous studies from other labs, we would get the same results. The mean age for the 4-year-olds was 4.0 years (SD = 0.22 years; range = 45.5–53.0 months). See **Table 1**.

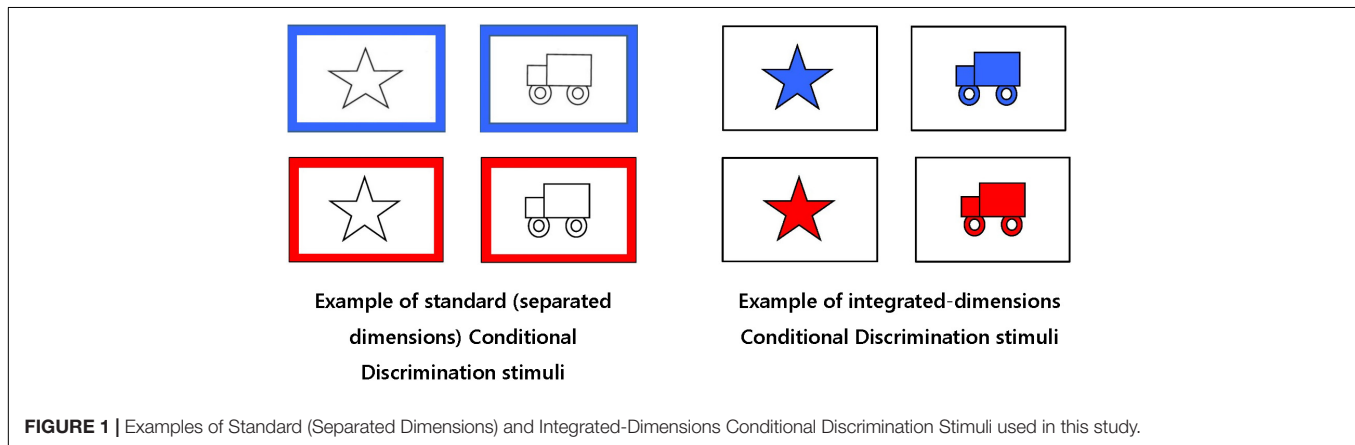
A priori power analyses using G*Power 3.1.9.2 (Faul et al., 2007) indicated that a total of 34 participants (11–12 per group) would provide 80% power to detect a medium effect size of 0.35.

Materials

A child sat directly across from the experimenter at a table measuring 76 × 76 × 55 cm. The child was seated in a child-sized chair (36 × 30 × 36 cm) and the experimenter was seated on a stool (20 × 39 × 23 cm). Two rectangular wooden boxes open at the top, each measuring 12.5 × 8.6 × 3.7 cm, served as the containers where the reward was hidden. These boxes were identical in appearance. At the base of one of the boxes on the inside was a marble well. The marble well held the marble in place to prevent the child from guessing the marble's location based on the sound of the marble rolling around. The stimulus cards served as the boxes' lids. The 12 cards for each condition measured 13.3 × 9.6 cm

TABLE 1 | Age, number, and sex of children in each group.

Condition	Age range	Mean age	SD age	#Male	#Female	N
Integrated	2.8–3.3 years	3.07 years	0.16	6	6	12
Separated	2.8–3.3 years	3.05 years	0.16	6	6	12
Separated	3.8–4.4 years	4.03 years	0.22	9	3	12



each and were laminated. For the integrated condition, the 12 stimulus cards displayed a star or a truck that was either blue or red on a white background. For the separated condition, the shapes (star or truck) were white outlined in black and the border of the cards was either blue or red. See **Figure 1**.

Marbles (1.5 cm diameter) of different colors and patterns served as the reward. When children found a marble, they could put it in our marble maze and watch as the marble soared down ramps and spun through turnstiles. The marble maze (see **Figure 2**) stood 28.0 cm tall and was on a flat platform measuring 20.0 × 15.5 cm. A plastic, transparent jar (6.5 × 6.5 × 11 cm) was used to display the trove of marbles a child had found.

Testing Procedure

First, the experimenter showed the child where to sit. The parent/guardian was given the option of sitting directly behind the child or watching from outside the testing room through a one-way mirror.

At the outset of testing, the experimenter told the child they were going to play a game and asked the child to cover his or her eyes like in the game “Peekaboo” (“*I am going to bring out a surprise. Can you cover your eyes, like this?*” [the experimenter showed child]). While the child’s eyes were closed and covered, the experimenter placed a marble in the marble well inside one of two boxes; this was the done underneath the table, out of sight even to the parent. Then the experimenter covered each box with a stimulus card (one showing a truck, the other showing a star) and placed the two boxes on the table, one to the left and one to the right, both equidistant from the child, and within the child’s reach. The left-right locations of the correct stimulus card was varied in the same pseudo-random sequence for all sessions (see below). The child sat, eyes covered, waiting in anticipation. The experimenter then announced: “*You can open your eyes now. I have hidden a surprise for you under one of these cards. Can you guess which one?*”

The child was encouraged to choose a card and lift it to see if the surprise was hidden beneath. If a child chose the correct card, the experimenter cheered exuberantly



and encouraged the child to retrieve the hidden marble. The child was then presented with the marble maze and shown how to place the marble in it, to the enormous delight of the child.

If the child chose the wrong card, the experimenter said disappointedly, “*Oh no, it wasn’t there.*” The experimenter then lifted the correct card and showed the child where the reward had been hidden and said, “*It was here, see? Let’s try again. You’ll find it next time!*” The child was thus given feedback on each trial and the experimenter either cheered happily or showed the child what the correct choice had been and encouraged the child to find the marble next time. At no point did the experimenter ever explicitly state that the marble was under the truck or star or state the conditional rule. In Block 1, all the cards contained blue. The marble was always hidden under the truck card. The right-left location of the stimulus cards was pseudo-randomly varied across trials (Truck: Left, L, Right, L, R, R, L, R, L, L, R, R – repeated as long as needed up to a maximum of 36 trials). Participants never saw the reward being hidden and were never explicitly told the rules of the game. To find the reward they had to deduce the rule governing where it would be. As the right (R) and left (L) locations of the stimuli were randomly varied, always reaching right or left as a strategy did not lead to success. Six consecutively correct trials were required to pass the block and move on to the next one.

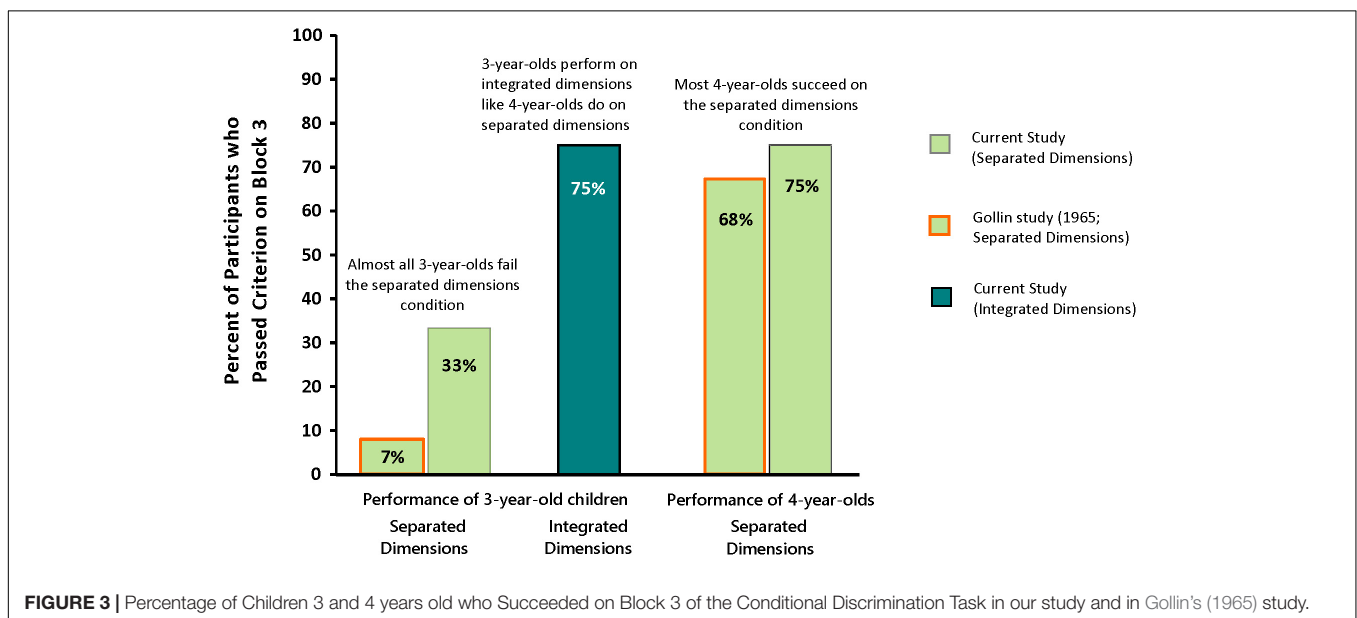
In Block 2, all cards contained red. Here, the marble reward was always hidden under the star stimulus card. Again, the right-left location of the stimulus cards was pseudo-randomly varied (Star: L, R, R, L, R, L, L, R, L, R, R, L – repeated up to a total of 3 times [36 trials]). The child needed to pick correctly on six trials in a row to pass criterion and move on to the final block.

On the first and second block, a child did not need to pay attention to color to choose correctly. The truck was always the correct choice for Block 1 and the star was always the correct choice for Block 2. There was no need to integrate color and shape information; attending to shape alone was sufficient.

In Block 3, cards containing blue or red were randomly intermixed over trials. On any given trial, both cards either contained red or blue. Again, the truck was the correct choice when both cards contained blue and the star was correct when both cards contained red. The following pseudo-random order indicates which color was presented on which trial and whether the correct choice was presented on the right or left: Blue + Left, Red + Right, Red + Left, Blue + Left, Red + Right, Blue + Right, Blue + Right, Red + Left, Blue + Right, Red + Right, Blue + Left, Red + Left. This was repeated as long as needed up to a maximum of 36 trials. As with Blocks 1 and 2, the criterion for passing Block 3 was six correct trials in a row.

The criterion for passing a block was 6 correct responses in a row within 18 trials. The choice of 18 trials was based on the work of Gollin and Liss (1962), who used 16–20 as their cut-off for Block 3 in their CD testing, after which the experimenter stepped in to aid the child in picking the correct stimulus. For each age X task group, we analyzed the number of trials needed to succeed on 6 trials in a row as well as the percentage of children who did so in 18 trials or less. We let children continue to try to figure out the CD rule after 18 trials, but only considered a child as having succeeded on a block if 6 correct trials in a row occurred within 18 trials or less.

Experimenter 2 (CDW) was blind to our hypothesis and predictions while testing the children. Experimenter 1 (DSL) was not blind to our hypothesis. Videos were taken of a random subset of those sessions where a parent gave permission (about 30% of sessions). The videos were reviewed by the senior author (AD) to check that children were treated comparably in both conditions and by both testers. Each experimenter also viewed the others’ tapes. AD noticed differences during practice and corrected them and would not approve the testers for testing until she was fully satisfied that they were doing each detail correctly and comparably. CDW and DSL each



tested 50% of the children of 3 years in each condition. For children 4 years of age, DSL conducted 67% of the testing and CDW 33%.

RESULTS

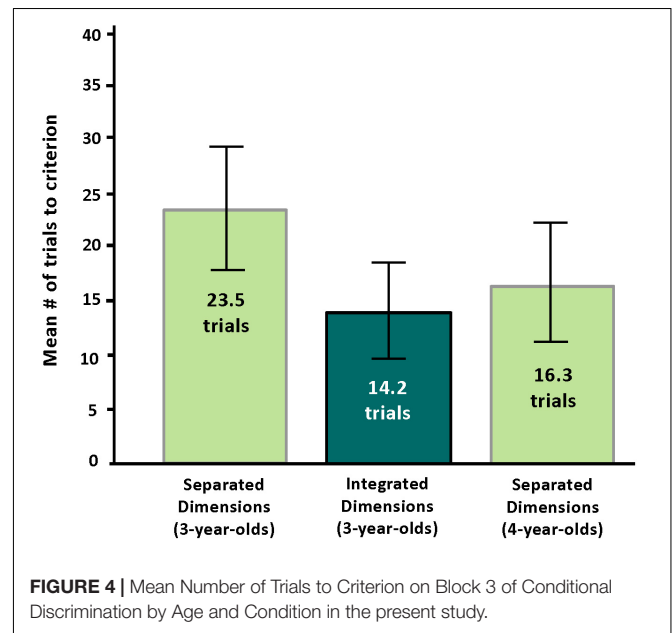
Tester was not significantly related to any dependent variable for any test of our hypotheses, nor were gender or tester X gender, so those variables were dropped from further statistical tests. Analysis of variance (ANOVA) was used to test all hypotheses except for hypotheses concerning the binary variable of pass/fail, for which Fisher's exact test was used.

Prediction 1: We turn first to our principal prediction, that children only 3 years old would succeed on CD with integrated dimensions. We operationalized "success" as performance by 3-year-olds on CD with integrated dimensions that is comparable to 4-year-olds on CD with separated dimensions. The percentage of 3-year-olds passing Block 3 of the integrated condition of CD (75%) was identical to the percentage of 4-year-olds passing Block 3 of the canonical version of the task (i.e., separated dimensions: 75%). See **Figure 3**. Children of 3 years took an average of 14.2 trials (SD = 7.8) to pass Block 3 when color and shape were integrated in the stimuli. Children of 4 years took an average of 16.3 trials (SD = 9.5) to succeed on the standard CD task when color and shape were separated in the stimuli. The number of trials needed to pass Block 3 was not significantly different between the two groups [$F(1,22) = 0.36$, NS]. Indeed, if anything, the number of trials was slightly lower for 3-year-olds with integrated dimensions than for 4-year-olds with separated dimensions. See **Figure 4**. We conclude that Prediction 1 was confirmed.

Prediction 2: We predicted that 3-year-olds would perform better on the integrated-dimensions condition than on the separated-dimensions condition. The percentage of 3-year-olds passing the integrated condition of CD (75%) was greater than the percentage of 3-year-olds passing the separated condition of CD (33%; Fisher's Exact Test, $p = 0.05$). See **Figure 3** above. Children of 3 years tested with separated dimensions (where color was a property of the border) took an average of 23.5 trials (SD = 9.8) to pass Block 3, whereas those tested with integrated dimensions (where color was a property of the truck or star) took an average of only 14.2 trials (SD = 7.8) to pass Block 3. That difference is significant: $F(1,22) = 6.71$, $p < 0.02$ (effect size: 0.23). See **Figure 4**. We conclude that Prediction 2 was confirmed.

Prediction 3a: We had predicted that we would replicate findings (Gollin, 1965) that 3-year-olds tested on CD with separated dimensions fail. Only 33% of the 3-year-olds tested with separated dimensions passed Block 3. Thus, most children of 3 years tested with the canonical version of CD failed (67%).

Prediction 3b: We predicted that when presented with the canonical CD task with separated dimensions, we would replicate previous findings (Gollin, 1965) that 4-year-olds succeed. Our results show that 75% of 4-year-olds tested on CD with separated



dimensions succeeded. This is similar to what Gollin (1965) found, which was that 68% of 4-year-olds succeeded. See **Figure 3**.

Prediction 3c: Lastly, we predicted that in all conditions and at both ages, children would succeed on Blocks 1 and 2 as these blocks are fairly easy. In both Blocks 1 and 2, color was irrelevant because on all trials both cards contained the same color (Block 1: Blue; Block 2: Red). Indeed, all children tested, regardless of age or condition, passed Blocks 1 and 2. Children of 3 years tested in the integrated condition took 10.2 and 13.3 trials respectively to pass Blocks 1 and 2. Children of 3 years tested in the canonical separated-dimensions condition took 12.5 and 16.0 trials respectively to pass Blocks 1 and 2. Children of 4 years tested in the canonical separated-dimensions condition took an average of 10.3 and 11.2 trials respectively to pass Blocks 1 and 2. We conclude that all three sub-components of Prediction 3 were confirmed.

As an aside, when Block 1 begins children have no idea which shape is correct. When Block 2 begins children have had experience over 10–12 trials on average where the truck was always the correct choice and the star was never correct. On Block 2 that reverses, now the star always indicates where the reward is hidden and the truck never does. Since Block 2 requires a reversal, we expected that it would take children a bit longer to perform consistently correctly on Block 2 than Block 1. It did take children slightly longer, but the difference in the number of trials to criterion in Blocks 1 and 2 was never significant in either condition or at either age: for 3-year-olds on separated dimensions: $F(1,10) = 1.19$, NS; for 3-year-olds on integrated dimensions: $F(1,10) = 2.98$, NS; for 4-year-olds on separated dimensions: $F(1,10) = 0.42$, NS. These results are controlling for gender; without controlling for gender the F -values are even lower.

Our results provide evidence that integrating the dimensions allows children to perform CD at a level roughly 12–18 months

ahead of when most had previously thought possible. Like others before us, we found that 3-year-olds fail and 4-year-olds succeed at CD presented the canonical way with color and shape separated on the stimulus cards (Gollin and Liss, 1962; Gollin, 1965; Rudy et al., 1993; Andrews et al., 2012).

DISCUSSION

We hypothesized that children of only 3 years are capable of conditional, if-then reasoning, but they do not seem to mentally integrate dimensions that are not properties of the same object. That is, when performing the CD task, they do not appreciate that the color in the background is telling them anything about which shape is the correct choice. On CD tasks children need to use the color shown to inform them which shape is correct (i.e., it is critical that they integrate color and shape information). We report here that when the dimensions of color and shape are integrated as part of the same stimulus, children of 3 years (12–18 months younger than previously reported) can use the value on one dimension (color) to indicate which value of the other dimension (shape) is correct, and thus succeed at CD. That is, they can deduce that red means the star is correct and blue that the truck is correct. When performing any task, one thing participants must do is determine which information in the environment is relevant and which is not. When color and shape are separated on the stimulus cards, 3-year-olds do not seem to comprehend that color is telling them anything about which shape is correct.

The current finding may be thought of as the flip-side of what we (Diamond et al., 2005) and Kloo and Perner (2005) found with the DCCS task. Children under 4 or 5 years typically fail to switch dimensions on DCCS. We hypothesized that if color and shape were not part of the same object, but instead if colorless shapes (black or white) were presented on cards with a background color, that children only 3 years old would be able to switch from sorting by color to sorting by shape or vice versa. Our hypothesis was confirmed (Diamond et al., 2005) and soon thereafter Kloo and Perner showed the same thing with colorless shapes and a color patch on each card. Children of 3 years can switch sorting dimensions when the dimensions are perceptually separate and not part of the same object.

The present findings together with those just cited for DCCS present the strongest evidence to date against several of the most prominent theories proposed for why 3-year-olds fail CD or DCCS. Evidently, 3-year-olds can grasp the hierarchical rule structure of the task (unlike Zelazo's influential Cognitive Complexity and Control – Revised [CCC-R] hypothesis; Zelazo et al., 2003), have sufficient memory (unlike Munakata's influential graded memory hypothesis; Morton and Munakata, 2002), and are capable of conditional, if-then reasoning (unlike Halford's influential hypothesis; Halford et al., 1998b; Halford et al., 2010; Andrews et al., 2012) since when superficial stimulus properties were changed, children of 3 years succeeded.

The perceptual bootstrap we provided through changing superficial properties of the stimulus cards removed neither the

need to grasp the embedded hierarchical rule structure, the memory demands of the task, nor the need for conditional “if, then” reasoning. Our study therefore suggests that scaffolding preschoolers' emerging conceptual skills by changing the way the stimuli look (perceptual bootstrapping) enables 3-year-olds to demonstrate if-then conceptual reasoning abilities long thought beyond their grasp.

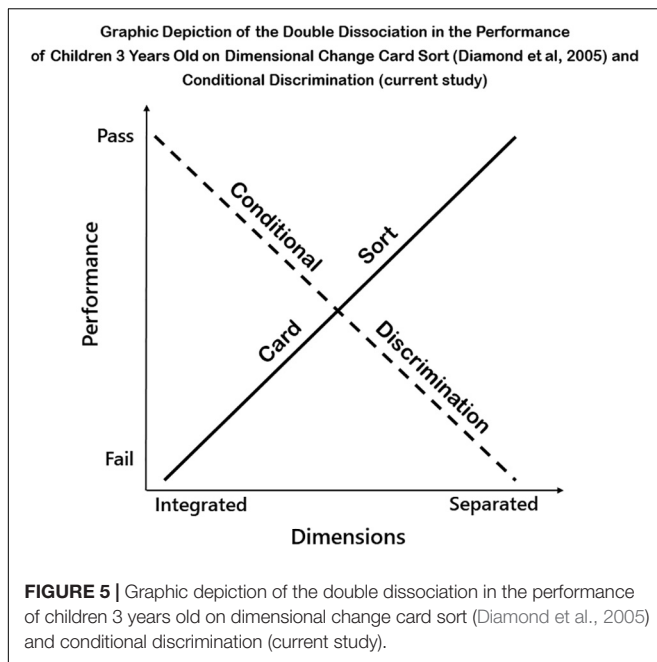
An alternative interpretation for our findings might be that success in the integrated-dimensions version of CD is due to simple associative learning, not conditional reasoning. Rather than learning that when the stimuli are blue, the truck is the correct choice, and when the stimuli are red, the star is correct, children might instead learn that “blue-truck” is correct and “red-star” is correct. Therefore, in Block 3 they simply scan for those two stimuli, and finding either, select it. An associative-learning interpretation, however, would have difficulty accounting for the findings that 3-year-olds needed an average of 8 trials and 4-year-olds needed an average of 10 trials before they started to consistently perform correctly in Block 3 (i.e., before their string of 6 correct responses in a row began). If it were simple associative learning, why did they need so many trials in Block 3? The first time, and every time, they saw a blue truck or red star they should have reached for that stimulus. Children should have been able to be consistently correct starting on Trial 1 of Block 3. Also, 2-year-olds, who are fully capable of associative learning, do not succeed at conditional discrimination, even with integrated dimensions. That is inconsistent with an associative-learning interpretation.

A more plausible alternative interpretation is kind of a linguistic interpretation. Perhaps children encode integrated stimuli as one word, e.g., red-star or blue-truck, like San-Francisco or South-Africa. Thus there is only one thing to remember for each condition. When the dimensions are separated, however, if the children even notice the color present in Blocks 1 and 2, they would need to hold 2 things in mind for each condition: star + red or truck + blue. Perhaps the latter puts too great a demand on their working memory. We cannot rule that out at present, though this explanation would not be applicable to the findings with DCCS.

The results reported here for conditional discrimination combined with those of Diamond et al. (2005) and Kloo and Perner (2005) on DCCS present a clear double dissociation between how integrating or separating the dimensions of color and shape affect the performance of 3-year-olds. What helps performance on CD hinders performance on DCCS and what helps performance on DCCS hinders performance on CD. **Figure 5** symbolically displays this dissociation.

While both the CD and DCCS tasks involve conditional reasoning, CD requires the integration of dimensions while DCCS requires the separation of those same two dimensions. Could it be that the way we have traditionally queried children has not made it possible for 3-year-olds to show evidence of their budding reasoning competencies? Perhaps what children need help with is in understanding which information in the environment is relevant and which is not.

We acknowledge that there are limitations to our study. Our sample sizes were small – 12 per group. Our results are clear, however, and our number of subjects sufficient to find significant



results. The sample sizes used here are quite comparable to those used by Kloo and Perner (2005) for testing separated versus integrated dimensions on the DCCS task. In one of their experiments, conditions were tested between-subjects (as here) and they had 12 participants per group (as here). In another of their experiments, conditions were tested within-subject with only nine participants per group. Another limitation is that we tested only one task (CD). It would have been more elegant to test the same children on both CD and DCCS. It would also have been more elegant to test each 3-year-old on both versions of CD, but we learned in using that design with DCCS that there was spillover from the easier condition to the more difficult one when tested within-child (i.e., children did better on DCCS with integrated dimensions when that was tested second than when it was tested first; Diamond et al., 2005). Also, CD testing takes longer than DCCS testing; almost none of the children we tested would have sat through another session testing the other CD condition. Another limitation is that Tester 1 was not blind to our hypothesis or predictions, as she had helped design the study. It is thus possible that she might have subtly, unintentionally affected children's performance. Tester 2, however, was blind to the study's hypothesis and predictions during testing, and we found no effect of tester on any outcome variable and no significant interaction between tester and any variable. Finally, we only tested children of 4 years on the integrated dimensions version of CD. While this last point might look like a limitation, since others have shown that 4-year-olds succeed on the more difficult version of CD (separated dimensions) it seemed unnecessary to test 4-year-olds on the easier version of the task (integrated dimensions).

Our results are consistent with those of other studies that used other paradigms. Jarvik's (1956) study shows perhaps the most astonishing evidence. Many studies had shown that it takes chimpanzees over 100 trials to learn a simple visual

discrimination (e.g., choose the red or green stimulus) when the reward is just below the stimulus card in a shallow well. Jarvik varied whether the reward was hidden 0.1 cm below the stimulus card in a shallow well or whether it was taped to a depression in the underside of the stimulus card. He replicated the result that with the reward in the well just below the stimulus card it takes chimpanzees an average of 131 trials to learn a visual discrimination. However, Jarvik found that chimpanzees were able to learn visual discriminations in *only one trial* when the reward was attached to the underside of the stimulus.

Our lab has previously shown the importance of perceptual modifications in other studies. When rewards were physically connected to the stimulus objects (e.g., by Velcro or even a string some inches long), infants only 9–12 months old could successfully use the stimuli to guide them to learn a delayed non-matching rule (choose the stimulus that does not match the sample you were just previously shown; Diamond et al., 1999; Shutts et al., 2001). When the reward is not attached to the stimulus object, but in the well just below, as in the canonical delayed non-matching to sample task, toddlers cannot succeed until they are 18–21 months old (Diamond, 1990; Overman, 1990; Diamond et al., 1994).

DeLoache's lab has likewise found results consistent with this: They report that toddlers of 18–22 months are significantly more likely to retrieve a reward they saw hidden when it is hidden inside a piece of furniture than when it is hidden near the same piece of furniture (DeLoache and Brown, 1983). Toddlers of 21 months can find a hidden object if it is hidden inside one of four attractive containers but they cannot use those same attractive containers to inform them where to search when those containers are mounted on top of four identical plain boxes (DeLoache, 1986).

In conclusion, if children of 3 years can succeed at CD when color and shape are integrated as part of the stimulus, then they must be capable of if-then, conditional reasoning at some level. It does not appear to be their reasoning ability that is lacking but rather what seems lacking is their ability to appreciate what information is relevant. Children of 3 years seem to rely on perceptual information (physical characteristics of the stimuli) to guide them in appreciating that the value of one dimension (color) is informing them about which value of the other dimension (shape) is correct.

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 Behavioural Research Ethics Board of UBC, Ethics Review Board of the Vancouver Coastal Health Research Institute, and Human Ethics Review Board of the Vancouver School Board. Written informed consent to participate in this study was provided by the participants' legal guardian/parent.

AUTHOR CONTRIBUTIONS

DL actively contributed to all aspects of this study and manuscript preparation. CW contributed to recruiting and testing the participants, entering the data, and thinking about what the results meant. AD actively contributed to all aspects of this study and manuscript preparation, except testing participants. 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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The Gestures in 2–4-Year-Old Children With Autism Spectrum Disorder

QianYing Ye, LinRu Liu, ShaoLi Lv, SanMei Cheng, HuiLin Zhu, YanTing Xu, XiaoBing Zou and HongZhu Deng*

Child Development and Behavior Center, Third Affiliated Hospital of Sun Yat-sen University, Guangzhou, China

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Els Ortibus,
KU Leuven, Belgium

Reviewed by:

Bart Boets,
KU Leuven, Belgium
Eva Murillo,
Universidad Autónoma de Madrid,
Spain

*Correspondence:

HongZhu Deng
dengzh@mail.sysu.edu.cn

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Deficits in gestures act as early signs of impairment in social interaction (SI) and communication in children with autism spectrum disorder (ASD). However, the pieces of literature on atypical gesture patterns in ASD children are contradictory. This investigation aimed to explore the atypical gesture pattern of ASD children from the dimensions of quantity, communicative function, and integration ability; and its relationship with social ability and adaptive behavior. We used a semi-structured interactive play to evaluate gestures of 33 ASD children (24–48 months old) and 24 typically developing (TD) children (12–36 months old). And we evaluated the social ability, adaptive behavior, and productive language of ASD and TD children by using the Adaptive Behavior Assessment System version II (ABAS-II) and Chinese Communication Development Inventory (CCDI). No matter the total score of CCDI was corrected or not, the relative frequency of total gestures, behavior regulation (BR) gestures, SI gestures, and joint attention (JA) gestures of ASD children were lower than that of TD children, as well as the proportion of JA gestures. However, there was no significant group difference in the proportion of BR and SI gestures. Before adjusting for the total score of CCDI, the relative frequency of gestures without vocalization/verbalization integration and vocalization/verbalization-integrated gestures in ASD children was lower than that in TD children. However, after matching the total score of CCDI, only the relative frequency of gestures without vocalization/verbalization integration was lower. Regardless of the fact that the total score of CCDI was corrected or not, the relative frequency and the proportion of eye-gaze-integrated gestures in ASD children were lower than that in TD children. And the proportion of gestures without eye-gaze integration in ASD children was higher than that in TD children. For ASD children, the social skills score in ABAS-II was positively correlated with the relative frequency of SI gesture and eye-gaze-integrated gestures; the total score of ABAS-II was positively correlated with the relative frequency of total gestures and eye-gaze-integrated gestures. In conclusion, ASD children produce fewer gestures and have deficits in JA gestures. The deficiency of integrating eye gaze and gesture is the core deficit of ASD children's gesture communication. Relatively, ASD children might be

capable of integrating vocalization/verbalization into gestures. SI gestures and the ability to integrate gesture and eye gaze are related to social ability. The quantity of gestures and the ability to integrate gesture with eye gaze are related to adaptive behavior.

Clinical Trial Registration: www.ClinicalTrials.gov, identifier ChiCTR1800019679.

Keywords: autism spectrum disorder, gesture, language, adaptive behavior, social ability

INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder with heterogeneous manifestations mainly characterized by impairments in social interaction (SI) and communication, as well as the presence of restrictive and repetitive behaviors [Diagnostic and Statistical Manual of Mental Disorders-Fifth Edition (DSM-V); American Psychiatric Association, 2013]. The Centers for Disease Control and Prevention reported that the rate of ASD has increased to 1 in 54 (Matthew et al., 2020). Deficits and delays in gestures act as early signs of impairment in SI and communication (Lebarton and Iverson, 2016).

Gestures in Typical Development Children

From very early in life, expressive behavior is multimodal, and early behavioral coordination is refined and strengthened for communication over time (Iverson, 2010). In the first months of life, typically developing (TD) children can communicate with others non-verbally through gestures, vocalizations, and facial expressions (Russell, 2007). It has been found that even during the prelinguistic stage, over half of the TD children can coordinate gesture and vocalization during communication (Wetherby et al., 1988). At 12 months, most TD children produce their first meaningful word (Blackwell and Baker, 2002). Before using word-word combinations, TD children start to use gesture-word combinations (Guidetti and Nicoladis, 2008; Iverson, 2010). Between 12 and 18 months, productive language and gesture are generally mutually exclusive (Capone and McGregor, 2004). At the multi-word stage, TD children use verbalization as their principal means of communication (Wetherby et al., 1988). However, from toddlers to preschoolers, combinations of gestures and verbalizations become more closely, and these cross-modal combinations can promote the development of language. At school age, gesture-speech mismatch occurs in TD children. Gesture-speech mismatch combination is a general cognitive phenomenon that reflects the transitional learning state for both science and math concepts (Capone and McGregor, 2004). In adults, communicative interactions are multimodal. They communicate with others through complex, fluid, and rapid coordination between speech, altering facial expression, changing eyebrows or head position, and gestures (Iverson, 2010).

Previous studies have found that gestures and early language development are closely linked (Bates and Dick, 2002). The development of gesture predates and predicts change in children's language development (Iverson, 2010). For instance, Camaioni

et al. (1991) found that communicative pointing at 12 months was significantly and positively correlated with vocabulary size at 20 months. And the number of different meaning gestures at 18 months predicted vocabulary at 42 months (Rowe and Goldin-Meadow, 2009). Additionally, the onset of gesture + word combinations could predict the emergence of word-word combinations (Iverson and Goldin-Meadow, 2005; Iverson et al., 2008; Iverson, 2010). Moreover, a study discovered that the number of gesture + speech combinations at 18 months predicted sentence complexity at 42 months (Rowe and Goldin-Meadow, 2009).

According to Bruner's three earliest functions of communication, gestures can be divided into three categories: SI gestures, behavior regulation (BR) gestures, and joint attention (JA) gestures (Bruner, 1981). BR gestures are used to coordinate other people's actions in order to make them do something or stop doing something; SI gestures are utilized to attract or keep others' attention to oneself, with the purpose of initiating or maintaining the interaction with each other; JA gestures are used to attract others' attention toward an event, an object, a person, or a topic, and just for showing or sharing (Bruner, 1981; Watson et al., 2013). A previous study reported that in TD children, BR gestures (at the mean age of 6.09 months) and SI gestures (at the mean age of 8.42 months) emerged before JA gestures (at the mean age of 9.33 months) (Crais et al., 2004). In general, gestures of these three communicative functions are seen in most TD children by 12 months of age (Watson et al., 2013). Wetherby et al. (1988) found that TD children displayed BR gestures and JA gestures most frequently in the second year of life.

Gestures in Children With Autism Spectrum Disorder

Previous studies explored the quantity of gestures in children with ASD of different ages and found that the main manifestation of ASD children is the reduction of gestures (Shumway and Wetherby, 2009; So et al., 2015; Lebarton and Iverson, 2016; Özçalışkan et al., 2016). Additionally, some studies discussed the communicative function and the integration ability of gestures in ASD children.

However, the conclusions of studies about the communicative function of gestures in ASD children were not entirely consistent. In early childhood, some studies have found that ASD children used fewer gestures (i.e., pointing, showing) to direct JA compared to TD children and children with language delay (LD) (Franchini et al., 2018). Additionally, Clifford et al. (2007) detected that ASD children used proto-declarative showing less than TD children and children with developmental disorders or LD at 12–24 months. Still, there

was no difference in proto-declarative pointing, social gestures, and request gestures. However, another study found that infants with autism used fewer BR gestures, SI gestures, and JA gestures than TD children and children with other developmental disabilities at 15–18 months (Watson et al., 2013). In the pre-school stage, ASD children displayed fewer JA gestures than language-matched intellectual disability (ID) children or mental-age-matched ID children and showed fewer requesting gestures than mental-age-matched ID children; however, there was no difference in SI gestures (Mundy et al., 1990). These suggest that ASD children are less likely to use JA gestures, while the use of BR gestures and SI gestures is not determined.

Previous research on the integration of gesture and other communicative means in ASD children discovered that ASD children displayed deficits in integrating gestures and vocalization/verbalization compared to TD children or LD children (Parladé and Iverson, 2015; Choi et al., 2019). However, Shumway and Wetherby (2009) found no significant difference in the proportion of total acts coordinated with a vocalization, eye gaze, and gesture at the same time between ASD children, TD children, and children with developmental delays. And Heymann et al. (2018) found that ASD children were less likely to integrate JA behaviors (including gestures and eye gaze) and vocalization than TD children. Furthermore, a study analyzed the complex level of integrating different communicative forms (gesture, vocalization, and eye gaze) and found that the level of complexity in ASD children is significantly lower than TD children (Maljaars et al., 2011). To conclude, studies on the integration of vocalization/verbalization, eye gaze, and gesture in ASD children were contradictory. Notably, when exploring the integration of gestures and eye gaze, the above studies included other communicative means, such as smile, voice, and language. Therefore, gestures which only integrated with eye gaze may be missed. The current study coded and analyzed the integration of gesture and vocalization/verbalization, and the integration of gestures and eye gaze separately. It may help us understand the cross-modal coordination ability of gestures more accurately and comprehensively for ASD children.

Recent studies showed that gesture use might also play an essential role in the development of language for ASD children. Özçalışkan et al. (2016) found that the deictic gestures of 30-month-old children with ASD could predict the vocabulary 1 year later. Besides, the emergence of distal pointing was closely linked with the emergence of first words, and the onset of gesture + word combinations predates and predicts the onset of two-word combinations (Talbot et al., 2018). In adolescence, gesture use was positively associated with vocabulary for youths with ASD (Medeiros and Winsler, 2014).

Current Study

Previous studies have shown that the use of gestures was influenced by the variation of culture (Kita, 2009; Kwon et al., 2017). However, most of the current research works on ASD children's gestures are based on Western-cultural conventions. This study was designed to explore the atypical

gesture pattern of ASD children from the dimensions of quantity, communicative function, and integration ability based on the Chinese-cultural convention; and its relationship with social ability and adaptive behavior.

MATERIALS AND METHODS

Participants

TD Group

Typically developing children were recruited through the internet or the Outpatient Department of Child Health Care at the Third Affiliated Hospital of Sun Yat-sen University in the corresponding period and met the following inclusion criteria: (1) Age 12–36 months. (2) The native language is Mandarin. (3) Parents of 12–18-month-old participants were asked to complete the Infant-Toddler Checklist (ITC; Wetherby and Prizant, 2002), and the results of the ITC must be “typical skills.” Parents of 18–36-month-old participants were asked to complete the Autism Behavior Checklist (ABC; Krug et al., 1978), and the total score of the ABC must be lower than 31. (4) All participants were confirmed as TD by two experienced developmental-behavioral pediatric specialists. The exclusion criteria are as follows: Developmental disorders, for example, ASD, ID, language disorder, etc.

ASD Group

All participants were recruited through the Child Developmental and Behavior Center of the Third Affiliated Hospital of Sun Yat-sen University from November 2018 to January 2020 and met the following inclusion criteria: (1) Age 24–48 months. (2) The native language is Mandarin. (3) All participants were diagnosed with ASD by two specialists in developmental-behavioral pediatrics using the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2003) and the Autism Diagnostic Interview-Revised (ADI-R; LeCouteur et al., 2003) following the DSM-V (American Psychiatric Association, 2013) criteria. The exclusion criteria are as follows: (1) Other developmental disorders, such as ID, language disorder, etc. (2) Genetic conditions associated with autism, for example, Rett syndrome, Fragile X syndrome, and tuberous sclerosis.

All participants were Chinese people. This research was approved by the Institutional Review Board at the Third Affiliated Hospital of Sun Yat-sen University and informed consent was obtained from the parents of all participants.

There are 33 children in the ASD group and 24 children in the TD group. All ASD children have received behavior intervention about 20–28 hours per week. There was no significant difference between the ASD and TD groups regarding gender, mother's educational attainment levels, and father's educational attainment levels. The mean age of the ASD group was older. Compared to the TD group, the scores of social skills, conceptual skills, practical skills in ABAS-II, the total score of ABAS-II, and the total score of CCDI were significantly lower in the ASD group. The characteristics and inferential statistics of participants are shown in **Table 1**.

TABLE 1 | Characteristics and inferential statistics of children by group.

		ASD (<i>n</i> = 33) M(SD)	TD (<i>n</i> = 24) M(SD)	<i>df</i>	$\chi^2/t/Z$	<i>p</i>
Gender	Male	26	17	1	0.474	0.491
	Female	7	7			
Age (months)		34.18 (6.67)	23.79 (7.33)	55	5.568	<0.001**
Mother's Educational Attainment Levels	Bachelor degree below	14	8	2	0.940	0.625
	Bachelor degree	15	14			
	Master degree or above	4	2			
Father's Educational Attainment Levels	Bachelor degree below	18	8	2	2.525	0.283
	Bachelor degree	13	14			
	Master degree or above	2	2			
ADOS	Communication and Social Interaction in model 1	13.92 (4.64)				
	Communication and Social Interaction in model 2	16.88 (3.83)				
ABAS-II	Social skills score	35.15 (17.12)	72.00 (17.93)	55	−7.867	<0.001**
	Conceptual skills score	55.85 (28.21)	99.88 (44.67)	36.096	−4.251	<0.001**
	Practical skills score	80.42 (31.62)	130.33 (51.59)	35.386	−4.200	<0.001**
	Total score	214.61 (81.58)	351.75 (118.01)	55	−5.192	<0.001**
CCDI	Total score	202.82 (230.14)	472.38 (364.76)		−2.917	0.004**

ADOS, Autism Diagnostic Observation Schedule; ABAS-II, Adaptive Behavior Assessment System version II; CCDI, Chinese Communication Development Inventory; M, mean; SD, standard deviation; *df*, degree of freedom. ***p* < 0.01.

Tasks

Assessment of Diagnosis

In this study, the ADOS (Lord et al., 2003) and ADI-R (LeCouteur et al., 2003) were used for diagnosing ASD. We used the Chinese version of ADOS and ADI-R, which was revised by Professor Wu YuYu of Taiwan and authorized by Western Psychological Service.

Assessment of Communicative Gestures

A doctor who was not familiar with participants evaluated the participants' gestural communications during a semi-structured play interaction. The content and sequence of play interaction were adapted from the ADOS. There were three main contents: blowing bubbles, blowing balloons, and snacking. Two social situations were set up in every content to encourage the children to express their demands or to show and share, while two kinds of communication opportunities. Child-initiated interaction and reactive interaction were also set up in every social situation. Only one parent was allowed to be present during the play interaction. The whole process of play interaction was videotaped for about 10 min by an assistant. The camera ensured that the child's face and hands were recorded at the same time and the doctor's face and hands. (The content and sequence of play interaction are shown in **Supplementary Appendix 1**).

Adaptive Behavior Assessment System Version II (ABAS-II; Oakland and Harrison, 2008)

The infant version of the Adaptive Behavior Assessment System Version II (ABAS-II) was used to assess the adaptive behavior of children aged 0–6 years. It is divided into a parent questionnaire and a teacher questionnaire. The adaptive behavior of children is evaluated from three levels. The first level is the overall adaptive function; the second level contains three composite areas of adaptive function: conceptual skills, social skills, and practical skills; the third level includes 10 concrete skill areas: communication, pre-school function, self-management, leisure, SI, community adaptability, family life, health and safety, self-care, and motor skills. Parents of participants completed the ABAS-II, which was revised by Professor Li YuQiu of Zhuhai Campus of Beijing Normal University and authorized by the American company, PEARSON. The social skills score and the total score (original score) were used to evaluate the social ability and adaptive behavior of participants. The higher the score, the better the social ability and adaptive behavior.

Chinese Communication Development Inventory (CCDI; Tardif et al., 2008)

Chinese Communication Development Inventory (CCDI) is the Chinese version of the MacArthur Communicative Development Inventories (MCDI; Fenson et al., 2007), which is filled out by parents. CCDI is used to assess the early language development of children aged 8–30 months who speak Chinese (Mandarin or Cantonese). CCDI can also be used to assess older children with developmental disorders. There are two forms in CCDI: the infant form (Words and Gestures) and the toddler form (Words and Sentences). We used the toddler form of the Mandarin CCDI, which is divided into two sections: productive vocabulary

and sentence complexity. The total score (raw score) of these two sections was used to evaluate the productive language of participants. The higher the score, the better the productive language. The highest total score of the toddler form of Mandarin CCDI is 903 (Tardif et al., 2008). In this study, there were 23 ASD children over 30 months old; and their mean CCDI total score is 236.13, with a minimum of 0 and a maximum of 795. There were six TD children over 30 months old; their mean CCDI total score is 778.17, with a minimum of 739 and a maximum of 841. In other words, no participants who were older than 30 months had a total score of CCDI above the 50th percentile score of 30 months old (boy: 844, girl: 850; Tardif et al., 2008), and this allowed us to use the CCDI to evaluate the productive language of all participants.

Gestures Coding

All behaviors of children in videos were coded using NVivo 12 (Windows) Pro software according to the following definitions.

Gestures

First of all, according to the checklist of coding gestures (**Supplementary Appendix 2**), we marked all target gestures of children. Second, we determined whether those gestures were used to communicate with another person (e.g., through the use of eye contact, vocalization, postural shift, repetition, or other interactive behaviors; Shumway and Wetherby, 2009; Paradé and Iverson, 2015; Özçalışkan et al., 2016). We excluded hand movements that were not used for communication. For example, we excluded imitation gestures (Braddock et al., 2015), hand movements that involved direct manipulation of an object, and hand movements that were part of a ritualized game (it should be noted that we did not exclude the showing gesture with communicative function; So et al., 2015).

The Communicative Function of Gestures

According to communicative function, gestures were coded using three categories (Bruner, 1981; Watson et al., 2013): (1) BR gestures are used to regulate another person's behavior to get another person to doing something or stop doing something. (2) SI gestures are used to attract or maintain another person's attention to oneself to initiate or maintain interaction. (3) JA gestures are used to draw another person's attention to an object, event, person, or topic which only for sharing.

Integration Ability of Gestures

Temporal co-occurrence is defined as the duration of different communicative behavioral overlaps at any time point. Vocalization: children's voices, such as vowel sound, laugh, cry, and squeal. Verbalization: single and multi-word spoken utterances. Vocalization/verbalization that was purely imitative (i.e., words repeated immediately after being spoken by another person) or not directed to another person were excluded (Shumway and Wetherby, 2009; Paradé and Iverson, 2015). With regard to any temporal co-occurrence between gesture and vocalization/verbalization, gestures were coded using these categories: vocalization/verbalization-integrated gestures and gestures without vocalization/verbalization integration.

Eye gaze is defined as the visual attention children paid directly to another person's eye region (Shumway and Wetherby, 2009; Paradé and Iverson, 2015). The eye region is defined as follows: In the horizontal direction, from the leftmost corner of the left eye to the rightmost corner of the right eye, and in the vertical direction, the area between the lower side of the eyebrow and the middle of the nose (He et al., 2019). The procedures of coding eye gaze: (1) Code the visual range of the child: We defined the visual range as within $\pm 20^\circ$ of the child's forward gazing direction. (2) Code the position in the relationship between child's visual range and doctor's eye region: We defined eye gaze behavior as that the visual range of the child can intersect with the eye region of the doctor (Figure 1). We defined the $\pm 20^\circ$ range based on previous research. Humans can pay visual attention to things inside the $\pm 20^\circ$ range around the facing direction, despite the direction the head faced. In contrast, they may choose to move their head when they pay visual attention to things outside of that range (Hachisu et al., 2018). Regarding whether there was any temporal co-occurrence between gesture and eye gaze, gestures were coded using these categories: eye-gaze-integrated gestures and gestures without eye-gaze integration.

Reliability

All videos were randomly assigned to two research assistants who were blind to group allocation. Two research assistants received coding training before coding the video separately. Approximately 20% of the participant videos were randomly selected and were double coded to calculate interrater agreement. The reliability of gestures coding between the two research assistants was estimated using an intraclass correlation coefficient (ICC) using an absolute agreement definition. The ICCs for the quantity of each gestures category (single measures) were as follows: total gestures (ICC = 0.940,

$p = 0.000$), BR gestures (ICC = 0.949, $p = 0.000$), SI gestures (ICC = 0.852, $p = 0.000$), JA gestures (ICC = 0.842, $p = 0.000$), gestures without vocalization/verbalization integration (ICC = 0.993, $p = 0.001$), vocalization/verbalization-integrated gestures (ICC = 0.936, $p = 0.000$), gestures without eye-gaze integration (ICC = 0.853, $p = 0.000$), and eye-gaze-integrated gestures (ICC = 0.989, $p = 0.000$).

Research Index

We utilized the relative frequency and the proportion of gestures in each category as research indexes. The conversion method was as follows: (a) the relative frequency of gestures: dividing the quantity of gestures in each category by the duration of videos in seconds separately and then multiplying by 600 to get the rate per 10 min of gestures in each category and (b) proportion of gestures: dividing the quantity of gestures in each category by the quantity of total gestures separately to get the proportion of gestures in each category.

Data Analysis

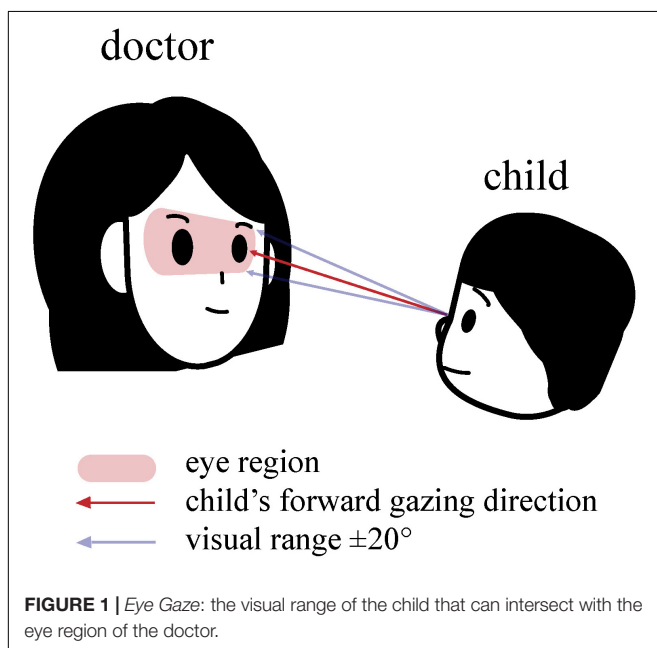
The analysis software used was SPSS Statistics version 23 (IBM Corp., Armonk, NY, United States), and the alpha was set at 0.05. Before conducting the data analysis, we detected the normal distribution of all data using normality tests. The total score of CCDI and some indices of gesture (the relative frequencies and the proportions of SI gestures and JA gestures, the relative frequency of eye-gaze-integrated gestures) were non-normally distributed, and the other variables were normally distributed. Therefore, Chi-square tests were conducted to explore the differences in gender and parents' educational attainment levels between the TD and ASD groups. *T*-tests were used to analyze the differences in age and the ABAS-II between the two groups. Non-parametric statistics (Mann-Whitney tests) were utilized to explore the differences in the CCDI between the two groups. After the logarithm transformations of the gestures indexes with non-normal distribution, analysis of variance (ANOVA) and analysis of covariance (ANCOVA, corrected for the total score of CCDI) were used to explore the differences in gestures between ASD and TD groups. Pearson correlation analysis was utilized to test the correlation between gestures, social ability, and adaptive behavior. Furthermore, partial correlation analysis was used to control the total score of the CCDI to study the associations between gestures, social ability, and adaptive behavior.

RESULTS

Considering that there were differences in the total score of CCDI between the ASD group and TD group, we use both ANOVA and ANCOVA (corrected for the total score of CCDI) when exploring the differences in gestures between groups.

The Quantity of Gestures

Whether we use ANOVA [$F_{(1,56)} = 43.2801$, $p^1 < 0.001$] or ANCOVA [corrected for the total score of CCDI, $F^2_{(1,55)} = 26.841$, $p^2 < 0.001$], we found that the relative



frequency of total gestures in ASD group was lower than that in TD group (Table 2).

The Communicative Function of Gestures

Whether we use ANOVA or ANCOVA, we found that the relative frequency of BR gestures [$F^1_{(1,56)} = 33.346, p^1 < 0.001$; $F^2_{(1,55)} = 20.136, p^2 < 0.001$], the relative frequency of SI gestures [$F^1_{(1,56)} = 9.453, p^1 = 0.003$; $F^2_{(1,55)} = 4.451, p = 0.040$], the relative frequency [$F^1_{(1,56)} = 17.111, p^1 < 0.001$; $F^2_{(1,55)} = 10.083, p^2 = 0.002$], and the proportion [$F^1_{(1,56)} = 8.416, p^1 = 0.005$; $F^2_{(1,55)} = 4.913, p^2 = 0.031$] of JA gestures in the ASD group were significantly lower than those in the TD group, while there was no significant difference in the proportion of BR gestures and SI gestures among groups ($p > 0.05$) (Table 2 and Figure 2).

The Integration Ability of Gestures

We found that the relative frequency of gestures without vocalization/verbalization integration [$F^1_{(1,56)} = 31.710, p^1 < 0.001$] and the relative frequency of vocalization/verbalization-integrated gestures [$F^1_{(1,56)} = 6.022, p^1 = 0.017$] in ASD group were significantly lower than that in TD group when we used ANOVA. While we utilized ANCOVA to adjust for the total score of CCDI, we found that only the relative frequency of gestures without vocalization/verbalization integration was significantly lower [$F^2_{(1,55)} = 22.644, p^2 < 0.001$]. Moreover, when we used ANOVA or ANCOVA, we found no significant difference among ASD and TD groups in the proportion of vocalization/verbalization-integrated gestures and gestures without vocalization/verbalization ($p > 0.05$).

Regardless of utilizing ANOVA or ANCOVA, we found that the relative frequency of eye-gaze-integrated gesture [$F^1_{(1,56)} = 41.840, p^1 < 0.001$; $F^2_{(1,55)} = 26.581, p^2 < 0.001$] and the proportion of eye-gaze-integrated gesture [$F^1_{(1,56)} = 28.864, p^1 < 0.001$; $F^2_{(1,55)} = 19.726, p^2 < 0.001$] in the ASD group were significantly lower than that in TD group. And ASD group showed a higher proportion in gestures without eye-gaze integration than the TD group [$F^1_{(1,56)} = 28.864, p^1 < 0.001$; $F^2_{(1,55)} = 19.726, p^2 < 0.001$]. Besides, we found no significant difference among ASD and TD groups in the relative frequency of gestures without eye-gaze integration ($p > 0.05$) (Table 2 and Figure 3).

The Relationship Between Gestures and ABAS-II

We found some statistically different gestural indexes between ASD and TD groups by utilizing ANCOVA. Those indexes might reflect the deficiency of ASD children's gestural communications most. Therefore, for reducing the number of calculated correlations, we only analyzed the correlation between ABAS-II and those gestural indexes. Besides, considering that the "proportion" indexes (i.e., the proportion of gestures without eye-gaze integration and the proportion of eye-gaze-integrated gestures) are merely complementary to each other, we only choose the proportion of eye-gaze-integrated gestures.

In ASD group, social skills score in ABAS-II was positively correlated with the relative frequency of SI gestures ($r = 0.368, p = 0.035$) and eye-gaze-integrated gestures ($r = 0.375, p = 0.032$); the total score of ABAS-II was positively correlated with the relative frequency of total gestures ($r = 0.401, p = 0.021$) and eye-gaze-integrated gestures ($r = 0.411, p = 0.017$). In TD group, the scores of ABAS-II were not significantly correlated with gestures ($p > 0.05$ for all) (Table 3).

When we controlled the total score of CCDI, the scores of ABAS-II were not significantly correlated with gestures in ASD group ($p > 0.05$ for all). In TD group, the social skills score in ABAS-II was positively correlated with the relative frequency of JA gestures ($r = 0.439, p = 0.036$) when controlling the total score of CCDI (Table 4).

DISCUSSION

The present study aimed to explore the differences in gestures between ASD children and TD children in different productive language levels. As expected, we found an atypical gestures pattern of ASD children from the dimensions of quantity, communicative function, and integration ability.

The Quantity of Gestures

We found that ASD children had lower scores of CCDI compared to TD children. And no matter that we corrected for the total score of CCDI or not, we found that ASD children displayed fewer gestures than TD children. ASD children have impairments in SI and social communication, and their communication deficits are not limited to spoken language but also gesture (Iverson et al., 2017). In other words, in the early development of life, ASD children produce fewer gestures than TD children regardless of their productive language. Similarly, Mastrogiuseppe et al. (2015) found that the quantity of gestures produced by ASD children (chronological age range 30–66 months) was significantly lower than in TD children.

The Communicative Function of Gestures

Before and after controlling for the total score of CCDI, we found that ASD children used less BR, SI, and JA gestures than TD children, and the proportion of JA gestures in ASD children was significantly lower. The differences in the relative frequencies of BR, SI, and JA gestures between ASD and TD groups might be due to the overall differences in gesture productions. And the possible explanation for the lower proportion of JA gestures is that JA gestures are related to more complex triadic interactions. For example, ASD children might need to coordinate attention between themselves, the doctor, and objects/location/event at the same time when using JA gestures (e.g., pointing to the bubble to let the doctor notice the bubble's location). In TD children, dyadic interaction with another person forms in the first 3 months of life, and dyadic interaction with object forms in the first 6 months of life. At 9–12 months, TD children begin to coordinate the two types of dyadic interactions to form triadic interactions (Bard, 2016). These complexities of triadic interactions might

TABLE 2 | Descriptive and inferential statistics for gestures of children by group.

			ASD (<i>n</i> = 25) M(SD)	TD (<i>n</i> = 12) M(SD)	<i>df</i> ¹	<i>F</i> ¹	<i>p</i> ¹	<i>df</i> ²	<i>F</i> ²	<i>p</i> ²
Quantity	Relative frequency	Total gestures	21.90 (8.07)	38.45 (10.94)	1,56	43.280	<0.001**	1,55	26.841	<0.001**
	Proportion (%)	Total gestures	21.90 (8.07)	38.45 (10.94)	1,56	43.280	<0.001**	1,55	26.841	<0.001**
Communicative function	Relative frequency	BR gestures	16.54 (5.19)	28.54 (10.30)	1,56	33.346	<0.001**	1,55	20.136	<0.001**
		SI gestures	2.66 (2.35)	4.38 (2.56)	1,56	9.453	0.003**	1,55	4.451	0.040*
		JA gestures	2.73 (3.72)	5.52 (2.98)	1,56	17.111	<0.001**	1,55	10.083	0.002**
		Proportion (%)	2.73 (3.72)	5.52 (2.98)	1,56	17.111	<0.001**	1,55	10.083	0.002**
	Proportion (%)	BR gestures	78.21 (15.76)	72.77 (10.39)	1,56	2.167	0.147	1,55	0.989	0.325
		SI gestures	11.56 (8.50)	11.81 (7.02)	1,56	1.088	0.302	1,55	0.248	0.621
Integration ability	Relative frequency	JA gestures	10.23 (11.28)	15.42 (9.65)	1,56	8.416	0.005**	1,55	4.913	0.031*
		Gestures without vocalization/verbalization integration	10.05 (5.69)	20.21 (7.95)	1,56	31.710	<0.001**	1,55	22.644	<0.001**
		Vocalization/verbalization-integrated gestures	11.84 (7.11)	18.15 (12.23)	1,56	6.022	0.017*	1,55	2.117	0.151
		Gestures without eye-gaze integration	12.22 (5.09)	11.90 (5.25)	1,56	0.054	0.816	1,55	0.057	0.813
	Proportion (%)	Eye-gaze-integrated gestures	9.37 (6.73)	26.55 (10.25)	1,56	41.840	<0.001**	1,55	26.581	<0.001**
		Gestures without vocalization/verbalization integration	46.28 (22.73)	55.75 (22.81)	1,56	2.401	0.127	1,55	3.052	0.086
		Vocalization/verbalization-integrated gestures	53.72 (22.72)	44.25 (22.81)	1,56	2.406	0.127	1,55	3.057	0.086
		Gestures without eye-gaze integration	58.52 (21.10)	31.82 (14.19)	1,56	28.864	<0.001**	1,55	19.726	<0.001**
		Eye-gaze-integrated gestures	41.48 (21.10)	68.18 (14.19)	1,56	28.864	<0.001**	1,55	19.726	<0.001**
		Gestures without eye-gaze integration	58.52 (21.10)	31.82 (14.19)	1,56	28.864	<0.001**	1,55	19.726	<0.001**
		Eye-gaze-integrated gestures	41.48 (21.10)	68.18 (14.19)	1,56	28.864	<0.001**	1,55	19.726	<0.001**

BR, behavior regulation; SI, social interaction; JA, joint attention; M, mean; SD, standard deviation; *df*, degree of freedom.

¹Analysis of variance.

²Analysis of covariance corrected for the total score of CCDI.

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

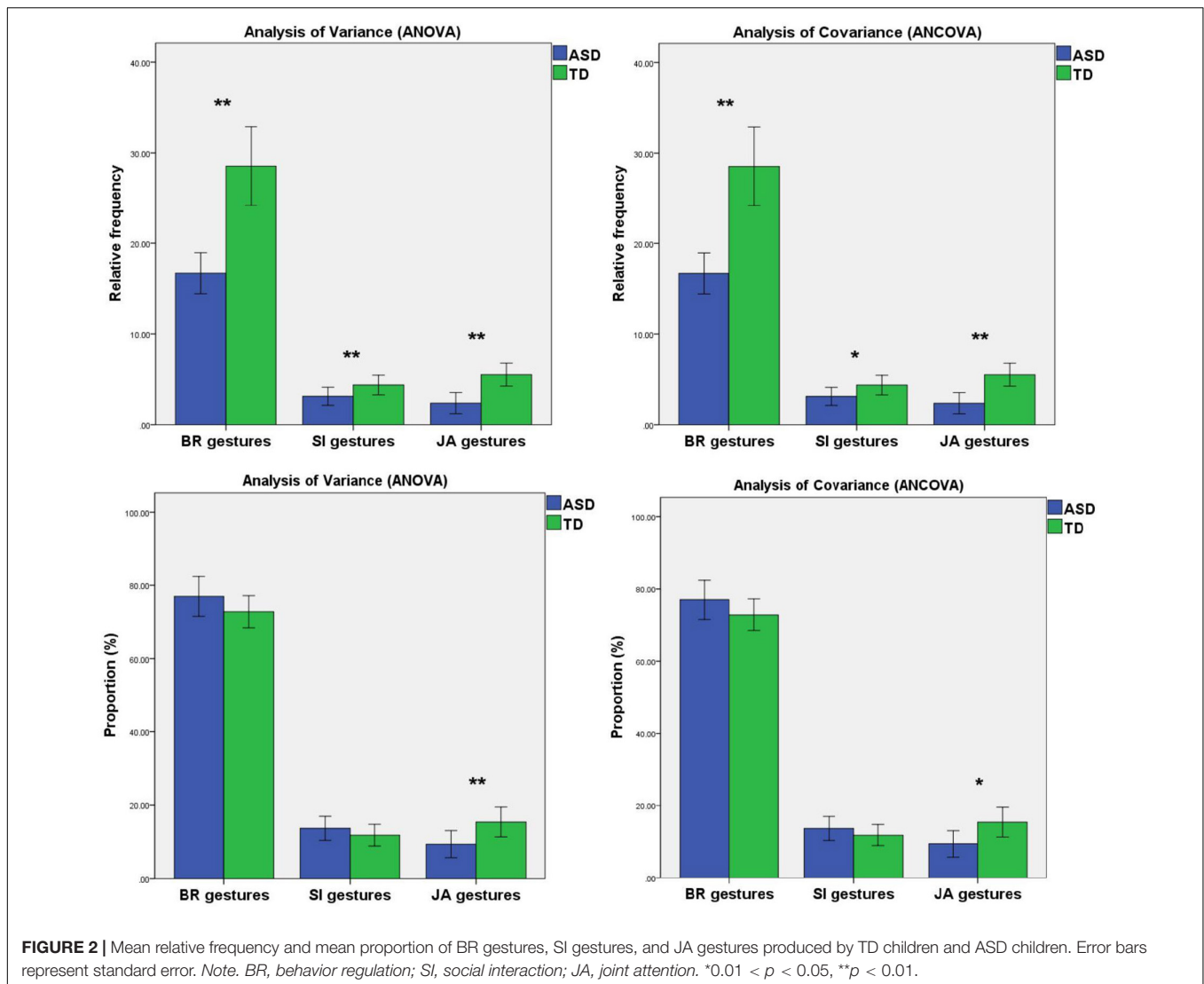


FIGURE 2 | Mean relative frequency and mean proportion of BR gestures, SI gestures, and JA gestures produced by TD children and ASD children. Error bars represent standard error. Note. BR, behavior regulation; SI, social interaction; JA, joint attention. * $0.01 < p < 0.05$, ** $p < 0.01$.

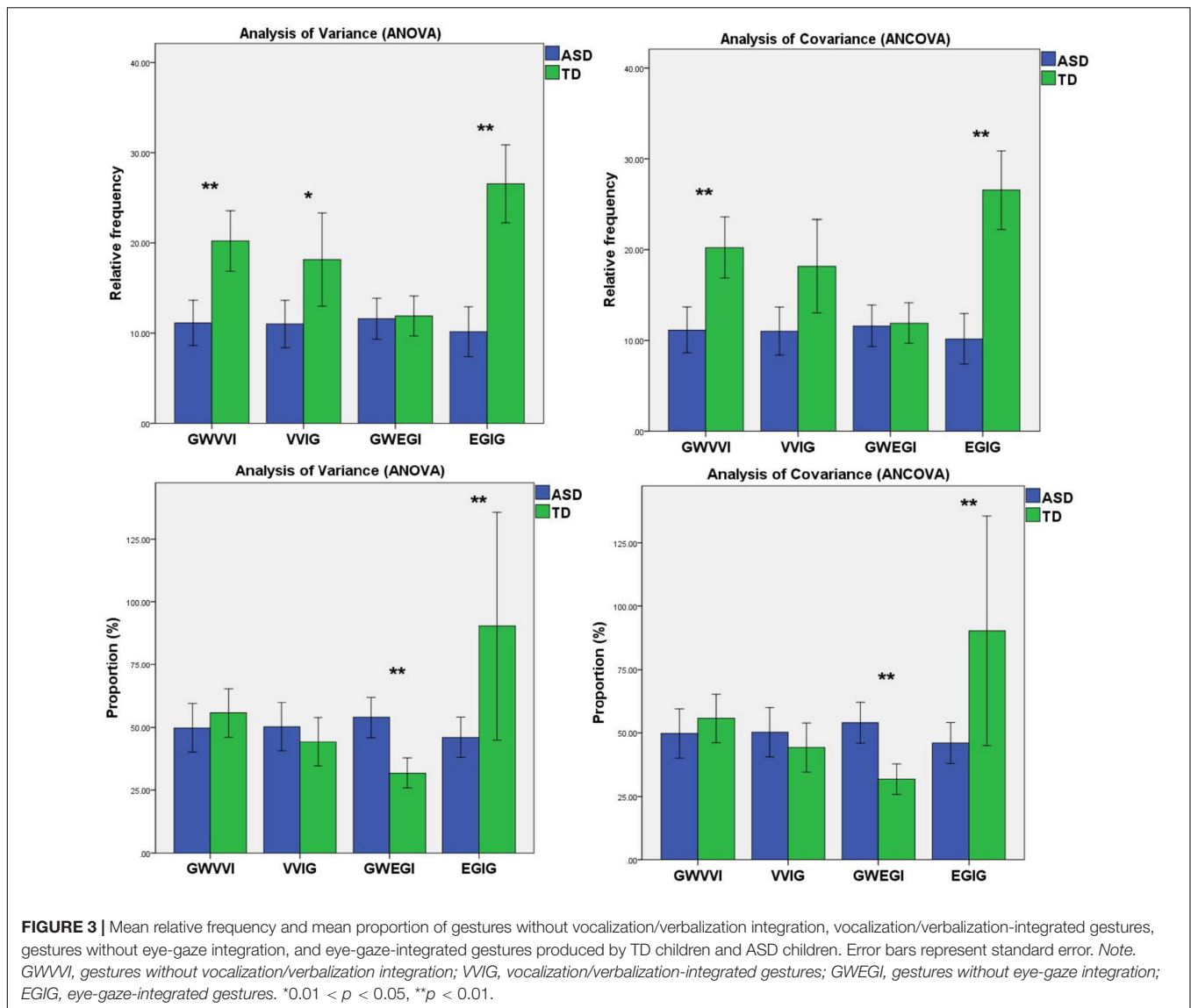
explain the deficits of ASD children in the use of JA gestures. Other studies have reported reduced triadic gestures in ASD children. For example, Watson et al. (2013) coded retrospective home videotapes and found that ASD children use fewer JA gestures at 9–12 and 15–19 months. In a prospective study of infant siblings at high-risk and low-risk ASD infants, Franchini et al. (2018) reported that initiations of JA were impaired from 12 months in ASD children, especially in the use of gestures (i.e., showing and pointing). Significantly, there was no difference in the proportion of SI gestures between ASD and TD children. One interpretation of this finding is that the limited semi-structured play situations might not have effectively triggered SI gestures from the children. Therefore, we may have underestimated the use of SI gestures in TD children.

The Integration Ability of Gestures

Before adjusting for the total score of CCDI, we found that ASD children were less likely to integrate gesture and vocalization/verbalization than TD children. However, after

correcting for the total score of CCDI, we found that the ability of ASD children to integrate gesture and vocalization/verbalization was no different from TD children. Recently, Murillo et al. (2020) found that there is no difference in the proportion of gesture + vocalization combinations between ASD children and language-matched TD children. Previous studies have shown that gestures will be combined with speech temporally and semantically when children enter the two-word stage of language development (Sowden et al., 2008). This suggests that the development of spoken language and gestures is concurrent. From the results of this study, we believe the reduction in the integration of gesture and vocalization/verbalization may merely be a potential sign of LD. ASD children might be capable of integrating vocalization/verbalization with gestures.

Conversely, no matter that we corrected for the total score of CCDI or not, ASD children were worse at integrating gesture and eye gaze than TD children. Likewise, Murillo et al. (2020) suggested that ASD children did not integrate gaze with gestures as TD children did, regardless of their productive vocabulary.



This indicates that eye gaze is closely related to the functional use (i.e., integration ability) of gestures. Previous studies have found that individuals with ASD have already experienced difficulties in social orientation in their infancy. Compared to TD individuals, individuals with ASD demonstrate decreased attention to socially relevant stimuli. In particular, they have a deficiency in processing the facial information of other people, as well as in establishing and maintaining eye contact (Guillon et al., 2014). We believe that the deficiency in the ability to integrate gestures with eye gaze seen in ASD children might be the core feature of their social impairment on the level of gestures.

The Relationship Between Gestures and Social Ability

For ASD children, the better social ability, the more SI gestures, and the better ability to integrate gesture and eye gaze. SI gestures are used to attract or maintain another person's attention to

oneself to initiate or maintain interaction (Watson et al., 2013). Thus, we decided that in terms of gestures, communication by SI gestures manifests better social ability in ASD children. Besides, previous studies have found that ASD children had impairment in facial perception; they reduced fixation on faces and eye region (Klin et al., 2002; Rice et al., 2012). The ability of facial perception of ASD children was related to their social ability (Klin et al., 2002; Mcpartland et al., 2011; Parish-Morris et al., 2013). Combined with the above results, we believe that the ability to integrate gesture and eye gaze in ASD children might be able to reflect their social ability.

The Relationship Between Gestures and Adaptive Behavior

Previous studies have shown that ASD children had deficits in adaptive behavior (Mouga et al., 2015; Bradshaw et al., 2018). The improvement of adaptive behavior is one of the crucial outcomes

TABLE 3 | Correlation between gestures and ABAS-II.

			Social skills score in ABAS-II				Total score of ABAS-II			
			ASD (n = 33)		TD (n = 24)		ASD (n = 33)		TD (n = 24)	
			r	P	r	P	r	p	r	p
Quantity	Relative frequency	Total gestures	0.328	0.062	0.322	0.125	0.401	0.021*	0.305	0.148
Communicative function	Relative frequency	BR gestures	0.266	0.135	0.201	0.347	0.339	0.054	0.211	0.323
		SI gestures	0.368	0.035*	0.262	0.216	0.315	0.074	0.282	0.182
		JA gestures	0.223	0.213	0.371	0.075	0.304	0.085	0.231	0.278
		Proportion (%)	0.167	0.354	0.245	0.249	0.229	0.141	0.128	0.551
Integration ability	Relative frequency	JA gestures	0.100	0.578	0.037	0.864	0.203	0.200	0.128	0.550
		Gestures without vocalization/verbalization integration	0.375	0.032*	0.364	0.080	0.411	0.017*	0.274	0.194
	Proportion (%)	Eye-gaze-integrated gestures	0.065	0.721	0.027	0.899	0.098	0.586	0.010	0.965

BR, behavior regulation; SI, social interaction; JA, joint attention; ABAS-II, Adaptive Behavior Assessment System version II; r, related coefficient. *0.01 < p < 0.05.

TABLE 4 | Partial correlation between gestures and ABAS-II.

			Social skills score in ABAS-II				Total score of ABAS-II			
			ASD (n = 33)		TD (n = 24)		ASD (n = 33)		TD (n = 24)	
			r	p	r	P	r	p	r	p
Quantity	Relative frequency	Total gestures	0.283	0.117	0.096	0.662	0.334	0.062	0.013	0.954
Communicative function	Relative frequency	BR gestures	0.253	0.162	−0.058	0.794	0.342	0.055	−0.116	0.597
		SI gestures	0.324	0.071	0.145	0.510	0.228	0.210	0.156	0.477
		JA gestures	0.145	0.427	0.439	0.036*	0.164	0.369	0.291	0.177
		Proportion (%)	0.095	0.605	0.389	0.066	0.098	0.594	0.289	0.181
Integration ability	Relative frequency	JA gestures	0.113	0.536	−0.114	0.604	0.253	0.163	−0.026	0.906
		Gestures without vocalization/verbalization integration	0.321	0.073	0.171	0.436	0.317	0.078	−0.013	0.954
	Proportion (%)	Eye-gaze-integrated gestures	0.029	0.876	−0.120	0.587	0.034	0.855	−0.194	0.376

BR, behavior regulation; SI, social interaction; JA, joint attention; ABAS-II, Adaptive Behavior Assessment System version II. r, related coefficient. *0.01 < p < 0.05.

of ASD intervention (Zachor and Ben-Itzhak, 2017). However, only a few studies preliminary discussed the relationship between adaptive behavior and gestures. For example, Kjellmer et al. (2012) found that for ASD children, non-verbal communications seem to be related to adaptive behavior, and Stamper et al. (2010) found that ASD children's deficits in gestural communication are related to adaptive behavior. Importantly, this study explored the relationship between gestures and adaptive behavior from the aspects of gestures' quantity, gestures' communicative function, and gestures' integration ability. The results showed that in ASD children, the number of gestures and the ability to integrate gesture and eye gaze positively correlate with adaptive behavior. It is probably because when ASD children produce more gestures, they communicate with others more. Furthermore, when communicating by gestures, eye-gaze integration may make communicative behavior more natural and smooth so that ASD children can better adapt to social life.

However, when controlling the productive language, the correlations between gesture, adaptive behavior, and social ability disappeared in ASD children. It may indicate that the relationships between gesture, adaptive behavior, and social ability are influenced by productive language. Future studies should explore the role productive language plays on relationships between gesture, social ability, and adaptive behavior.

In the TD group, we found no significant relationship between gestures, social ability, and adaptive behavior. However, when we controlled productive language, the social ability was positively correlated with JA gestures. That is probably because the development of gestures in TD children is more closely related to language development (Goldin-Meadow and Alibali, 2013).

Limitation

By reviewing research, we can find that TD children's gestures and language develop rapidly in the second and third years after birth. Consequently, we enrolled 12–36 months TD children. And gestures and early language development are closely linked. It is necessary to consider the impact of language when exploring the difference in gesture patterns between ASD children and TD children. In some previous research on gestures of ASD children, the chronological age of TD children ranged from 12 to 36 months. After matching, they enrolled ASD children who are 1–2 years older than TD children (Mastrogioseppe et al., 2015; Özçalışkan et al., 2016, 2017). According to these research works, we enrolled 24–48 months ASD children to make the productive language between ASD children and TD children more comparable. In future work, we should use relevant assessments to match productive language development between the ASD and control groups. The semi-structured play situation may elicit communication strategies that are not operated by ASD children in naturalistic situations. In the future, we can investigate the pattern of ASD children's gestures in natural situations by coding family videos. Moreover, there are only TD children in the control group of this study. Children with other developmental disorders need to be included in future studies to ensure that the results are more specific. Last, in the associations

between gestures and ABAS-II, the significance level around $0.01 < P < 0.05$, which might be a consequence of the Type 1 error chance. In future studies, we should increase the number of participants and set up the significance level of $p < 0.01$.

CONCLUSION

We discovered the atypical gesture patterns of ASD children: (1) ASD children produce fewer gestures and have deficits in triadic interaction gestures (i.e., JA gestures). (2) The deficiency of integrating eye gaze and gesture is the core deficit of ASD children's gesture communication. Relatively, children with ASD might be capable of integrating vocalization/verbalization into gestures. Furthermore, we found that SI gestures and the ability to integrate gestures and eye gaze are related to the social ability. The quantity of gestures and the ability to integrate gestures with eye gaze are related to adaptive behavior.

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 Medical Ethics Committee of the Third Affiliated Hospital of Sun Yat-sen University. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

QY, XZ, and HD contributed to conception and design of the study. QY and SC organized the database. QY, LL, and HZ performed the statistical analysis. QY wrote the first draft of the manuscript. QY, SL, YX, and HD wrote the sections of the manuscript. All authors contributed to manuscript revision, and read and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsyg.2021.604542/full#supplementary-material>

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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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Play First Before Doing Your Exercise: Does Acting in a Game-Like Task Improve 5-Year-Olds' Working Memory Performance?

Christophe Fitamen^{1,2*} and Valérie Camos¹

¹Département de Psychologie, Université de Fribourg, Fribourg, Switzerland, ²Laboratoire de Psychologie Cognitive, UMR 7290, Université d'Aix-Marseille and CNRS, Marseille, France

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Fondation Asile des Aveugles,
Switzerland

*Correspondence:

Christophe Fitamen
christophe.fitamen@unifr.ch

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It has been shown that acting in a game-like task improves preschoolers' working memory when tested in a reconstruction task. The game context and the motor activity during the game would provide goal cues bringing support to the memory processes. The aim of the present study was to test this hypothesis by examining preschoolers' working memory performance in a game-like task compared to an exercise-like task, which offers less goal cues. In the present study, 5-year-olds had to maintain a series of fruits and vegetables while acting in a game-like task or remaining static during the same task presented in a school-exercise context (within-subject factor). Memory performance was tested either through oral recall or reconstruction of the series of memory items (between-subject factor). Despite the fact that memory performance did not differ between the two conditions (game vs. exercise) whatever the type of memory tests, performance was worst in the game-like than in the exercise condition when the exercise was presented first. No difference emerged between conditions when the game condition was performed first. This result suggests that preschoolers were able to take advantage of acting in the game-like condition to integrate some task requirements, which were beneficial for performing the exercise condition.

Keywords: working memory, preschoolers, action, game, goal cue

INTRODUCTION

Do your exercise first and then you can go playing! Any child has already heard this. We will not question here the fact of being rewarded for the effort during homework. On the contrary, we will see that starting with a fun activity, like a game, would allow children to benefit from it for the achievement of a following, more academic, activity. The hypothesis we tested in the present study is related to the maintenance of the goal during an activity. In previous studies, it has been shown that preschoolers' performance in inhibition and cognitive flexibility tasks is impaired when they cannot actively maintain the goal during the task (Marcovitch et al., 2007, 2010; Chevalier and Blaye, 2008; Yanaoka and Saito, 2017). However, it is possible to help preschoolers to effectively maintain the goal during the task by presenting them with meaningful contextual cues related to the goal to be pursued

(Towse et al., 2007; Chevalier and Blaye, 2009; Blaye and Chevalier, 2011). Alternatively, it has been shown that some motor activity like gestures provide support to cognitive performance. In the present study, we hypothesized that acting in a game-like context can provide meaningful goal cues because it joins the motor activity and contextual cues that would have a favorable impact on preschoolers' working memory performance. Before examining the role of goal cues in executive functions, we present the impact of motor activity in supporting working memory.

Working Memory and Influence of Motor Activity

According to some theoretical conceptions, working memory is part of executive control (Diamond, 2016), and in charge of the storing and processing of information at short term (Baddeley, 1986). Working memory is involved in learning processes such as reading (Cain et al., 2004; Cain, 2006), in text comprehension (Carretti et al., 2005), in arithmetical activities where it predicts subsequent success (De Smedt et al., 2009), in reasoning, and in all other high-level cognitive activities (Camos and Barrouillet, 2018, for a review). As a consequence, working memory capacity is an excellent predictor of academic success (Gathercole and Alloway, 2004), and measures of working memory capacity also provides a better prediction than does the assessment of IQ based, in part, on the assessment of general knowledge as reading skills and mathematics (Alloway, 2009; Alloway and Alloway, 2010). In addition, working memory capacity is a better predictor than socio-economic level (Barrouillet et al., 2008) and does not depend on knowledge acquired before school period (Alloway et al., 2004).

Recently, research in cognitive psychology has investigated the support that motor activity can provide to working memory, especially the role of gestures. For example, in solving additions, children with poorer working memory capacity use strategies such as counting on their fingers to compensate their difficulties (Geary et al., 2004). In the same vein, according to Cook et al. (2012), producing meaningful gestures could reduce the cognitive cost in young adults when they solve a mathematical problem. The help of gestures when solving a math problem has also been observed in children aged 9 and 10 years. More specifically, children exhibited better performance in solving a problem when they received task instructions to use gestures or when the experimenter herself used gestures compared to children who had neither seen gestures from the experimenter nor did gestures (Cook and Goldin-Meadow, 2006). In a study testing 10-year-old children and young adults, Goldin-Meadow et al. (2001) tested that the use of gestures reduced working memory load. Before solving math problems, children and adults were presented with a list of words or letters to be recalled after the problem solving. The authors found that when children and adults were able to use gestures during the problem solving, their recall performance of the memory list was better than when they did not use gestures. Hence, performing gestures during the concurrent task of problem solving would free up cognitive resources for the memory task (see also Goldin-Meadow, 2011). In the same vein, a study by So et al. (2012)

involving children aged 4 and 5 years provides evidence in favor of the usefulness of gestures in a verbal memory task. Children were involved in three different conditions of a verbal memory task. Children watched a videotaped narrator who recited a list of verbs and produced meaningful gestures who were iconic gestures, or children saw the narrator reciting the verbs and produced beat gestures simultaneously, or they watched the narrator reciting the verbs without any gesture. When children recalled the verbs after a 2 min delay, their performance was better in the first condition, the two others did not differ. Hence, gestures can help children in improving their performance either on a memory task or on a secondary memory task by freeing cognitive resources during the primary task.

Moreover, it has been shown that another type of motor activity, i.e., walking, can also help memory performance. In a study involving 9-year-old children and young adults, participants were asked to perform an auditory *n*-back (one-back to four-back) task. In this task, participants heard one letter at a time and, for example, in the case of a two-back, they must spot when a new letter is identical to the one presented two letters before. While participants were doing the *n*-back task, they either walked on a treadmill or remained seated (Schaefer et al., 2010). When walking on the treadmill, participants performed either at their preferred pace or at a slower pace than their usual walking speed. Both children and adults exhibited better performance in the *n*-back task when they freely choose their walking speed compared to the slower-pace or remaining-seated conditions. In addition to studies on the role of gestures, this study converges in showing that motor activities can provide support to memory performance in adults, school-age children, and even preschoolers.

In the present study, we would like to suggest that the benefit from the physical involvement of children in performing an activity results from the active maintenance of the task goal, because in many cases, this physical involvement is oriented toward the goal of the task.

Goal Maintenance by Enactment

Two studies previously conducted by Istomina (1975) and Bertrand and Camos (2015) highlight the importance of the link between performing an action that makes sense in relation to the pursued goal and the active maintenance of that goal. In an old experiment first published in Russian in 1948, Istomina (1975, for the English translation) tested long-term memory in children aged 3–7 years old under two experimental conditions. Children were engaged either in a condition where they remained static and which was akin to a school-exercise condition, or in a condition requiring a motor activity in which they had to take part in a role play, doing shopping. In the exercise condition, children had to listen attentively to the words said by the experimenter to orally recall them after a 60–90 s delay. In the shopping condition, lists of items were presented as a shopping list and children had to go to a toy shop and asked for the items to another child playing the merchant. Preschoolers showed better recall performance in the play than in the exercise condition. The author suggested that the shopping game context emphasized

the task goal (memorizing for further recall) through the provision of a pragmatic relevance to recall. Hence, the overall context of the task can provide some cueing that would help to maintain the task goal, resulting in improved memory performance. It is noteworthy that because walking is itself goal-directed, it could also play the role of a goal cue that would support goal maintenance in children. This last interpretation was raised by Bertrand and Camos (2015) in their study, in which the authors implemented a rather similar game situation to assess preschoolers' working memory. The task was also a shopping game in which 4- to 6-year-old children had to memorize lists of verbal items for further recall either immediately after the presentation of the items or after a delay. Preschoolers' working memory performance improved when they had to walk straight to a toy shop instead of waiting seated in the front of the shop during the same delay. Among their interpretations, the authors suggested that walking improved goal maintenance in preschoolers and led to the observed better recall performance, because it is a goal-oriented motor activity. To summarize, findings of these two studies suggest that when children's memory capacity is assessed in a goal-supportive context this can help improving preschoolers' memory. Moreover, where there is an enactment of a situation, this can provide some support to the goal maintenance leading to improved memory performance.

A recent study by Fitamen et al. (2019) brought further evidence to support this last hypothesis. In a computerized working memory task, 5-year-old children had to memorize lists of items while they watched an animation of either a schoolbag that symbolized the container of the memory items, or a non-meaningful rectangle. Moreover, in two further conditions, children had to follow with their finger the movement on screen of the schoolbag or the rectangle. Children exhibited their best recall scores when they had to track the schoolbag. Hence, the concomitance of contextual cues (the schoolbag) and the motor involvement (tracking) led to improvement in working memory performance. The present study aimed at testing the joint effect of contextual cues and action in improving working memory performance in preschoolers by extending this previous finding into a more natural setting, akin to the situations used in Istomina (1975) and Bertrand and Camos (2015).

The Present Study

In the present study, we enrolled 5-year-old children in two experimental conditions, one enriched in contextual cues and proposing an oriented motor activity toward the goal (game condition), the other presenting neither contextual cues nor motor activity (exercise condition). In the game condition, those children were involved in a role play of shopping where after memorizing a list of items they walked to a market stall. This condition was similar to Bertrand and Camos (2015). In the exercise condition, children were involved in an exercise situation more comparable to their everyday classroom exercises where they also had to memorize a list of words while sitting

in front of experimenters. This condition was comparable to exercise condition of Istomina (1975). We hypothesized that the joint effect of action and contextual cues on goal maintenance in these natural settings should improve memory performance and children should exhibit better recall scores in the game than in the exercise condition.

However, such a beneficial effect of the action and contextual cues on memory performance should occur only when the task does not in itself favor the goal maintenance. In Bertrand and Camos (2015) study as well as in some conditions of Istomina (1975), children performed a reconstruction test. Hence, during the testing phase, children were asked to collect the previously encoded fruits and vegetables in a box containing different elements. This type of tests could encourage goal neglect because children know since the beginning of the task that the items will be presented to them during the testing phase. They may not try to actively maintaining memoranda and the goal during the delay of retention. Moreover, it can be assumed that a reconstruction test can be carried out by appealing only to familiarity of memory traces stored in long-term memory (see Yonelinas, 2002, for a review; Malmberg, 2008) that is without having to actively maintain memory traces in working memory. Thus, the type of tests implemented at the end of a working memory task could impact the goal maintenance. For example, in a Stroop task (Kane and Engle, 2003) and in a card sorting task (Marcovitch et al., 2007), when the task required frequent reactivation of the goal (i.e., predominantly incongruent condition in the former and predominantly conflicting condition in the latter), errors decreased compared to conditions that did not require active goal maintenance (predominantly congruent condition in the former and redundant condition in the latter). Hence, the characteristics of the test can more or less call for goal maintenance. In a reconstruction test that in itself provides retrieval cues at test, goal can be more easily neglected than in an oral recall test in which children had to rely on active maintenance to produce the memory items.

To test this additional hypothesis, we manipulated the type of tests by proposing to 5-year-old children either a reconstruction test or an oral recall test. We expected better memory performance in the reconstruction than in the oral recall test, replicating the difference reported between recall and recognition tests (see Tiberghien and Lecocq, 1983, for a review), although the reconstruction test is situated between recognition and recall as it has to preserve the serial order unlike recognition test, but like recall test. Moreover, we hypothesized that goal neglect would occur in the task with the reconstruction test and not with the oral recall test. Under the reconstruction test, children should then perform better in a play condition that helps goal maintenance than in an exercise condition that did not provide any goal support. However, under the oral recall test, we should not observe any effect of the type of contexts (exercise or game) on children's working memory performance. Thus, we expected to observe an interaction between the type of tests (reconstruction vs. oral recall) and the type of contexts (exercise vs. game).

MATERIALS AND METHODS

Participants

Sixty-two 5-year-olds ($M_{age} = 4;11$, $SD = 0;4$, 30 girls) took part in the experience. The mother tongue was French for all children. The experiment took place at the children's school in a quiet location. The experiment was approved by the local ethics committee, and we gathered from the parents or legal guardians a consent form. Children gave also their consent orally before beginning the experiment.

Three children were excluded from the analyses. One was followed in occupational therapy, another in speech therapy, and a last one could not sufficiently maintain his attention during the second experimental condition making the task unworkable. This led to a final sample of 59 children, randomly assigned to the two tests (29 in reconstruction and 30 in recall test).

Material and Procedure

The design was adapted from Istomina (1975) and Bertrand and Camos (2015). The experiment had a mixed design with the type of tests (reconstruction vs. oral recall) as between-subject factor, and the type of contexts (exercise vs. game) as within-subject factor. The order of presentation of the two conditions of context was counterbalanced.

To assess the similarity of the two groups in working memory capacity, every child performed before the experimental conditions the Number Recall subtest of the K-ABC 2 with 3 series in each length ranging from 2 to 9 digits, except for length 8 with only one series (Kaufman and Kaufman, 1993). Testing stopped after three successive series not correctly recalled. In this subtest, each correctly recalled series gave 1 point, and the raw score was the sum of the point (maximum score = 22). Before the experimental session, we also assessed the distance each child can walk at her own pace in 4 s in one training trial and three test trials. The average distance walked on the test trials determined the walking distance in the game context (see below). The distance was hence adapted to each child (mean = 4 m and $SD = 1$ m).

Nine different experimenters were involved in the study, but only two intervened with each child. One experimenter was in charge of the encoding part while another experimenter took care of the recall part of the working memory task. Before starting the working memory task, the experimenter verified at the encoding that the child recognized each plastic item representing fruits and vegetables. The fruits and vegetables (banana, tomato, orange, lemon, and carrot) were selected to have French bi-syllabic names with high frequency (Lété et al., 2004), an early age of acquisition (in years, 1.58, 1.65, 1.62, 1.88, and 1.58, respectively; Alario and Ferrand, 1999), but also different shapes and colors to be easily distinguished from each other. Children had to memorize lists of 1–4 fruits and vegetables. Four series were presented in each length, a given item appearing only once in each series. However, each item was presented in several series, which prevents that recall relies only memory traces from long-term memory. Two lists of

memory series were created, one per condition of context (exercise vs. game) for each child. A trial started when the experimenter took one fruit or vegetable, named it and put it in a transparent tube-shaped bag narrow enough to keep items on top of each other, arranged in a single column, the child paying attention to the scene. The items were successively introduced in the bag at a roughly regular rate of one every second. When all the items of the series were in the experimenter's bag, the bag was hidden to the child's eyes. Then, after a 4-s delay, the child had to reproduce the series according to the conditions she was assigned to (see below for the description of the four different experimental conditions). The child proceeded to the next length if she produced perfect recall (i.e., correct fruits and vegetables in correct order) on at least one trial of a given length. Each child had to reproduce series of items in two different conditions (exercise vs. game). Children performed the two conditions in the same room.

For the *exercise condition with oral recall test*, the child stayed seated in front of two experimenters (one for encoding, one for recall) after the "encoding" experimenter's bag was hidden, and waited for an auditory signal heard after 4-s delay. At the signal, the "recall" experimenter opened an opaque box, placed between the child and the experimenters, and which contained the five different fruits and vegetables that were not visible to the child. Once the child has orally recalled an item, the "recall" experimenter took it from the box and put it in a transparent tube-shaped bag similar to the bag used for the encoding. The *exercise condition with reconstruction test* was similar, except that the box was opened in front of the child so that she could see and grab easily one by one the fruits and vegetables to reconstruct the memorized sequence. The child put herself the fruits and vegetables in her transparent tube-shaped bag during the reconstruction test.

In the two *game conditions* (*reconstruction* and *oral recall*), the child had to walk with their empty bag straight to the shopping stall after the presentation of the items and the signal of the experimenter in charge of encoding to "go ahead." After 4 s, the recall experimenter who played the merchant opened the box placed on the stall. The child performed the test depending on the condition (*reconstruction* or *oral recall*) in the same way as in the exercise conditions.

A span score was computed for each child in each condition. Each correctly recalled series (i.e., in which all the items were correctly placed in the order of presentation) counted as one-fourth, and the total number of fourths added (Smyth and Scholey, 1992; Barrouillet et al., 2009; Bertrand and Camos, 2015).

RESULTS

A first ANOVA was performed on the raw scores of the Number Recall subtest of the K-ABC 2 with the type of tests, the lists, and the order of presentation of the type of contexts as between-subject factors. All effects were non-significant, $ps > 0.10$. Importantly for the purpose of the present study, the two

groups of children that were randomly assigned to each condition of tests (reconstruction: mean = 7.0, SD = 2.2; oral recall: mean = 6.8, SD = 2.0) did not differ on the Number Recall task, $F(1,51) = 0.065$, $p = 0.80$, $\eta_p^2 = 0.001$.

A second ANOVA was performed on span scores with the type of contexts as within-subject factor, and the type of tests, the lists, the order of presentation of the context conditions as between-subject factors. The only significant effect was the interaction between the type of contexts and its order of presentation, $F(1,51) = 8.46$, $p = 0.005$, $\eta_p^2 = 0.142$. The values of p for the other effects were higher than 0.20. It should be noted that the interaction of interest between the type of tests and the type of contexts was non-significant, $F(1,51) = 0.456$, $p = 0.503$, $\eta_p^2 = 0.009$. To take into account individual differences, we added in a third ANOVA the score at the digit span task as covariable. The same pattern of findings emerged as in the previous analysis. As expected, the score at the digit span task had a significant effect on the recall performance of our main tasks, $F(1,50) = 25.70$, $p < 0.001$, $\eta_p^2 = 0.340$. Except this last effect, the only other significant effect was the interaction between the type of contexts and its order of presentation, $F(1,50) = 8.80$, $p = 0.005$, $\eta_p^2 = 0.150$. The interaction of interest between the type of tests and the type of contexts remained non-significant, $F(1,50) = 0.502$, $p = 0.482$, $\eta_p^2 = 0.010$. This absence of interaction was confirmed by the analyses comparing the type of contexts within each type of tests, $t(50) = 1.43$, $p = 0.16$ and $t(50) = 0.41$, $p = 0.68$ in recall and reconstruction tests, respectively.

To summarize, only the interaction between the type of contexts and its order of presentation accounted for the results observed on the span scores. Children starting with the exercise condition (mean = 2.32, SD = 0.5) had a significantly lower span score during the game condition (mean = 2.04, SD = 0.5) presented afterward, $t(50) = 3.04$, $p = 0.004$. However, working memory performance in children starting with the game condition (mean = 2.27, SD = 0.5) did not differ in the exercise

condition presented afterward (mean = 2.16, SD = 0.5), $t(50) = 1.18$, $p = 0.25$ (**Figure 1**).

DISCUSSION

In this study, our aim was to test the hypothesis that the joint effect of a highly meaningful context and a goal-oriented motor activity during a working memory task would influence children's ability to maintain the goal and improve working memory performance. Moreover, this aid would be favorable to preschoolers only in the case of a reconstruction test, which favors goal neglect, whereas this aid should not affect an oral recall test that encourages goal maintenance in children. If such a combined help of a highly meaningful context with a goal-oriented motor activity in a reconstruction test can effectively boost goal maintenance, then we should observe a beneficial effect on preschoolers' working memory performance. Our results did not support our hypothesis. First, type of contexts (game vs. exercise) and the type of tests (reconstruction vs. oral recall) did not affect memory performance, and no interaction was evidenced between these two variables. Only an interaction between the type of contexts and its order of presentation was significant. Children showed degraded working memory performance in the game condition when they started with an exercise condition. This detrimental effect was not observed when they started by the game condition followed by the exercise condition. The results thus appeared at odds with those of Istomina (1975) and Bertrand and Camos (2015), in which recall performance was improved in a game situation, which included both a goal cue and a motor activity, and this even with a reconstruction test. The results are also contradictory to Fitamen et al. (2019) who observed a benefit in 5-year-old children's working memory performance in a situation combining a goal-oriented motor activity in a

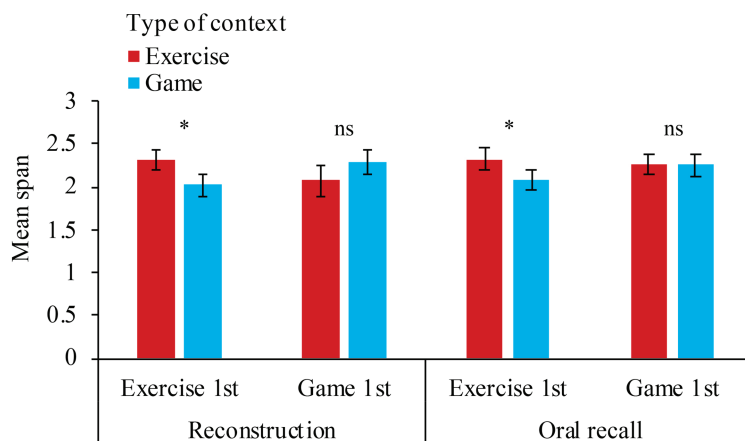


FIGURE 1 | Mean span as a function of the type of contexts (exercise vs. game), the order of presentation of the context conditions (exercise in 1st vs. game in 1st), and the type of tests (reconstruction vs. oral recall). Vertical bars represent SEs. Ns for non-significant difference on t -tests comparing the type of contexts in each pair of conditions and * for significant difference at $p < 0.05$.

meaningful context. In the following, we examined the reasons why such a discrepancy appeared compared to the two previous sets of studies.

To understand the discrepancy in findings and the lack of beneficial effect in the reconstruction test, one can examine the design of the encoding phase. Brown (1975) has shown that children of 5 years of age are able to get similar memory performance in reconstruction and oral recall tests, when the temporal order of to-be-memorized information is in direct correspondence with the representation of its spatial order during the encoding phase. Concretely, this happens when children had to memorize items presented as pictures in a retention array in the same (spatial) order as they appeared (temporally) in a story told at the same time (Brown, 1975, Exp. 2). On the contrary, when the encoding does not make the link between spatial and temporal representations possible, which means that items occupied scrambled spatial positions when the story was presented, children obtained better memory performance in the reconstruction test compared to the oral recall test (Brown, 1975, Exp. 1). In the present study, a direct correspondence between the temporal and spatial orders can be built up during encoding. Indeed, by using a thin transparent bag at encoding, children had access simultaneously to the temporal representation of the order (one fruit or vegetable per second placed in the bag) and the spatial representation of this order (by looking at the column of fruits and vegetables in the bag). Based on Brown's (1975) findings, our particular encoding condition can explain the absence of effect of the type of tests in the present study. This could also confirm that the link between temporal and spatial representations during encoding is critical in preschoolers, while this effect disappeared in older children (7–8 years of age) in study of Brown (1975, Exp. 1).

The present findings are also at odds with Fitamen et al. (2019) who reported a beneficial effect on working memory of the combination of contextual cues and action in 5-year-olds. Although the previous study and the present one shared the fact that the motor activity is related to the container of the items (the schoolbag in Fitamen et al., 2019; and the shopping stall in the present study), the main difference between the two studies is the implementation of the tasks. While we chose here a rather naturalistic setting akin to the daily activities of preschoolers (playing a shopping game, doing school-like exercise), Fitamen et al.'s (2019) task was computerized and presented on a tablet. Although further studies are required to examine in more details the divergence of findings, this discrepancy questions the transfer of effects observed in tablet to natural settings. In recent years, the use of tablets and computers to test young children became a norm and it provides several advantages for experimental psychology (e.g., better control of the conditions, collect of more fine-grained data). Nevertheless, the present study gave an example of how difficult it is to directly transfer knowledge from laboratory to classroom, and calls for more care when implications for practice are drawn from laboratory tests.

Finally, the absence of interaction effect between the type of contexts and the type of tests in the present study

contradicts the idea that favoring goal maintenance has a decisive impact in 5-year-olds' working memory performance, contrary to our hypothesis. Nonetheless, the interaction between the type of contexts and its order of presentation might indicate that performing a game situation first enabled children to effectively set the goal. Indeed, when the first condition is the game context, the context helps the goal identification thanks to the highly significant contextual characteristics of the game context (e.g., visual cues provided by the shopping stall, goal-oriented walk). The requirements of the memory task (e.g., remembering that the goal is to memorize, implementing maintenance strategies) can be transferred to the second (exercise) condition in which the goal was less salient. The grocery game condition, by giving a clearer meaning as to why memorizing shopping items (i.e., doing the shopping), could thus have served as a sort of tutorial that allow keeping the performance at the same level in the second (here exercise) condition. This tutorial effect could be beneficial thanks to contextualized learning. When performing the game condition first, children were engaged concretely in a meaningful activity. Then, they were able to transpose what they experienced in a living and concrete activity toward a more abstract activity, when doing the exercise as second condition. This enactment of the memorization situation is, moreover, one of the accounts suggested by Bertrand and Camos (2015) to explain the increase in working memory performance in a condition similar to the present game condition. On the contrary, working memory performance was reduced in the game condition when presented as second condition, for two reasons. First, children started with the exercise condition cannot benefit from the same kind of tutorial and contextualized learning as in the game condition, the goal of the task being less salient. Second, in the game condition, children had to process more information (e.g., understanding the story, looking at the shopping stall, and moving toward the stall), which could impair their memory capacity as their attentional resources need to be allocated to more information. This increased attentional demand added to the tiredness or weariness accumulated by the children during the first exercise condition may have been detrimental to working memory performance in the game condition when presented second. On the contrary, the high attentional demand induced by the game condition could have been adequately managed when this condition was presented first, and that attentional resources were still intact. In a follow-up study, the same type of context could be repeated within the same group of children (i.e., performing twice the game or exercise condition), to disentangle the effect of the condition from the potential effect of tiredness or weariness.

To conclude, the present study examined ways to improve working memory performance in preschoolers by providing contextual cues and motor activity. Contrary to laboratory testing condition, the implementation of the combination of contextual cues and motor activity did not benefit working memory performance in a more naturalistic setting. Nevertheless, the

presentation at first of the task as a game seems to provide some information to preschoolers that they can transfer in a second attempt, contrary to the presentation as an exercise. Further studies are needed to strengthen this result and examine its determinants.

DATA AVAILABILITY STATEMENT

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found at: <https://mfr.osf.io/render?url=https%3A%2F%2Fosf.io%2Fu89tm%2Fdownload>.

ETHICS STATEMENT

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

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AUTHOR CONTRIBUTIONS

CF and VC wrote the manuscript. CF prepared the **Figure 1**. Both the 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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Effect of Handwriting on Visual Word Recognition in Chinese Bilingual Children and Adults

Connie Qun Guan^{1,2*}, Elaine R. Smolen³, Wanjin Meng^{4*} and James R. Booth⁵

¹ Faculty of Foreign Studies, Beijing Language and Culture University, Beijing, China, ² Department of Psychology, Carnegie Mellon University, Pittsburgh, PA, United States, ³ Teachers College, Columbia University, New York City, NY, United States, ⁴ Institute of Psychology, Moral and Special Education, National Institute for Education Sciences, Beijing, China, ⁵ Department of Psychology and Human Development, Vanderbilt University, Nashville, TN, United States

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Beijing Normal University, China

*Correspondence:

Connie Qun Guan
qunguan81@163.com
Wanjin Meng
1085760333@qq.com

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In a digital era that neglects handwriting, the current study is significant because it examines the mechanisms underlying this process. We recruited 9- to 10-year-old Chinese children ($n = 24$), who were at an important period of handwriting development, and adult college students ($n = 24$), for both behavioral and electroencephalogram (EEG) experiments. We designed four learning conditions: handwriting Chinese (HC), viewing Chinese (VC), drawing shapes followed by Chinese recognition (DC), and drawing shapes followed by English recognition (DE). Both behavioral and EEG results showed that HC facilitated visual word recognition compared to VC, and behavioral results showed that HC facilitated visual word recognition compared to drawing shapes. HC and VC resulted in a lateralization of the N170 in adults, but not in children. Taken together, the results of the study suggest benefits of handwriting on the neural processing and behavioral performance in response to Chinese characters. The study results argue for maintaining handwriting practices to promote the perception of visual word forms in the digital age.

Keywords: handwriting, embodied cognition, N170, laterality, plasticity

INTRODUCTION

The development of the ability to write meaningful symbols was a major milestone in the development of human civilization. Handwriting serves to link auditory and motor routines with visual word processing, which is a hallmark for successful reading (Dehaene and Cohen, 2011). Early processing of visual word forms is constrained by the interaction with auditory and motor regions (Sekiyama et al., 2003; Wuerger et al., 2012; Callan et al., 2014), and the mechanism elicited by handwriting movement facilitates the auditory and motor integration of visual word forms (Longcamp et al., 2006; Guan et al., 2011; James, 2017).

Handwriting using Chinese characters appears to differ in several important ways from writing using an alphabetic system, such as that used in English. When handwriting Chinese, the individual needs to extract the visual-spatial features of the characters first. In contrast, for alphabetic words, phonological processing, such as mapping the letters corresponding to the phonemes, is more important. Giving up handwriting may affect how future generations learn to read (James and Engelhardt, 2012; Tan et al., 2013). Reducing handwriting instruction and practice may contribute significantly to difficulties in children's reading development (James, 2010; Guan et al., 2011; Tan et al., 2013) and overall writing skills (Daly et al., 2003; van Reybroeck and Michiels, 2018; Guan et al., 2019) in Chinese and Western languages.

Handwriting affects symbol learning by creating a network that includes both sensory and motor brain systems. James (2017) have demonstrated that the motor system creates variability (through handwriting in this case) in our perceptual world that enhances behavioral performance and serves to link brain systems into functional networks. In addition, a series of handwriting behavioral studies in both native English-speaking adults and Chinese beginning readers has suggested that handwriting Chinese characters focuses attention on stroke components (Guan et al., 2015) and facilitates orthographic recognition to aid reading acquisition among Chinese learners (Guan and Fraundorf, 2020; Guan et al., 2020). It may even be the case that drawing promotes Chinese children's cognitive ability in reading Chinese characters (Tan et al., 2013). A practical implication of these studies is that handwriting practice can be important parts of courses in Chinese to support more robust student learning of the spoken and written language.

The N170 is a component of the event-related potential (ERP) and is a neurophysiological indicator of early visual word recognition. Visual specialization for reading is revealed by the topography of the N170 ERP response (Maurer et al., 2005a). The N170 ERPs seem to represent a logographic processing strategy in visual word recognition (Simon et al., 2007). Selectivity of the N170 in the left hemisphere is also an electrophysiological marker for expertise in reading Chinese (Zhao et al., 2012) and Japanese (Maurer et al., 2008). However, whether handwriting experience enhances the N170 is unknown. We did not focus on other early visual ERP indicators (such as P1 and N1) because they are non-linguistic (Planton et al., 2013; Rothe et al., 2015). Focusing only on N170 modulation and the laterality effect is innovative, as previous relevant studies did not manipulate handwriting experience. Therefore, whether handwriting experience compared to other learning conditions might trigger this N170 modulation is unknown.

In summary, there is still controversy to what extent handwriting can promote the perception of words/characters. In particular, whether handwriting Chinese might promote visual word recognition more than visual perception or drawing is still unexplored. Moreover, there have been no direct studies comparing the role of handwriting in learning for children vs. adults.

The Current Study

The current study focuses on not only the difference between handwriting and viewing but also the difference between handwriting and drawing followed by Chinese recognition and drawing followed by English recognition. Specifically, we investigate whether the early neural mechanism of visual processing is different between the four learning conditions by examining the N170. The following research questions guide the present investigation:

- (1) What are the differences between the effect of handwriting and the effect of viewing characters in terms of individuals' behavioral and ERP responses?
- (2) What are the differences between the effect of handwriting and the effect of drawing followed by Chinese recognition in terms of individuals' behavioral and ERP responses?
- (3) What are the differences between the effect of drawing followed by Chinese recognition and the effect of drawing followed by English recognition in terms of behavioral and electroencephalogram (EEG) responses?
- (4) What is the difference in lateralization of the facilitative effect of handwriting between children and adults?

MATERIALS AND METHODS

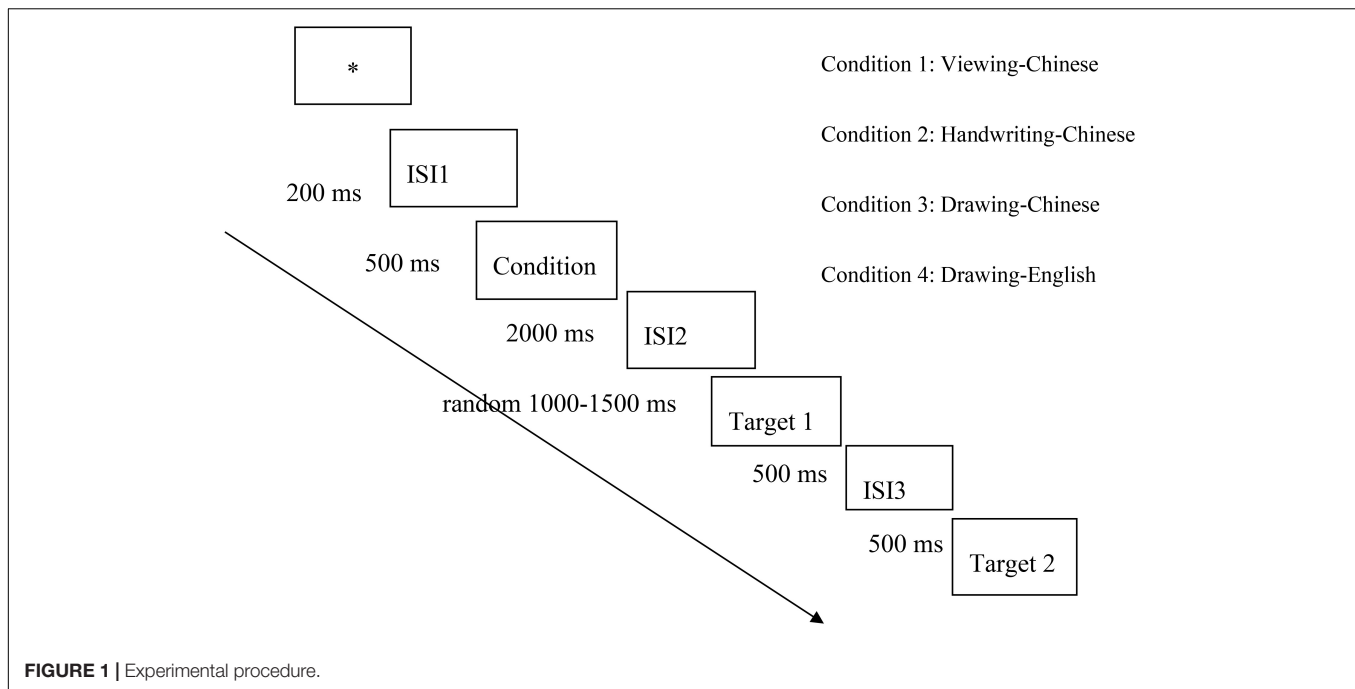
Participants

The University of Science and Technology Beijing (USTB) ethics committee approved the study. Parents of the children and the college students first signed the Informed Consent Form and then completed a background survey of developmental disorders and learning disabilities. After screening, 24 children (15 males, $M_{age} = 9.5$ years, $SD = 0.86$) in grades 3 and 4, who were at the significant period of handwriting development, participated in the experiment. Twenty-four undergraduates (eight males; $M_{age} = 19.8$ years old) from the USTB also participated in the experiment. All the participants were right-handed with normal or corrected-to-normal vision and no history of psychiatric or neurological disorders. Transportation and accommodations were reimbursed for participants who had to travel to the experiment site. The local participants were compensated 30 yuan (approximately \$4.50 US) per hour.

Materials

Study materials included Chinese characters and English words that were selected from the children's Chinese and English textbooks. These Chinese character materials have been used in previous studies (Guan and Fraundorf, 2020; Guan et al., 2020; Guan and Geva, under review); details about the selection process can be found in Guan et al. (2020). The materials included the prompt, target 1, and target 2. Chinese stimuli included 心, 乙, 人, 飞, 九, 儿, 口, 工, 日, 王, 十, and 田. Characters were selected for target 1 (32 in total) based on the following criteria: (1) high frequency (occur frequently in standard Chinese writing), according to the work of Chen and Shu (2001); (2) easy to embed in complex or compound characters; and (3) simple characters that contained either curved-line strokes or straight-line strokes. Target 2 comprised compound characters that contained the target 1 characters. Target 2 (32 in total) characters were chosen based on configuration (left-right, up-down, inside-outside) and familiarity. The characters-to-be-learned and the targets were counterbalanced with characters' curving or straight features. The number of strokes for characters of target 2 was always higher than that for the target 1 characters. See **Appendix 1** for detailed Chinese stimuli.

The English materials comprised all capital letters or words. During the learning conditions, the stimuli were H, F, I, T, E, L, O, C, Q, and U, six straight-line letters and four curved letters. Target 1 (32 in total) contained 26 capital letters. Target 2 (32 in total) comprised words containing 4–6 of these capital letters. The



words chosen were judged to be known to all participants, which controlled for the effect of familiarity. See **Appendix 2** for the English stimuli. The judgment task was the same for both Chinese and English: to decide whether target 1 was embedded in target 2.

Procedures

This study used a within-subject design. The independent variables were four conditions [handwriting Chinese (HC), viewing Chinese (VC), drawing Chinese (DC), and drawing English (DE)]; the dependent variables were behavioral performance [accuracy (ACC) and response time (RT)] and the ERP component (N170).

The experiment used four learning conditions. The first learning condition was VC, under which participants only needed to view the blue stimulus of Chinese words and then respond to the judgment target task by making a binary decision on whether target 2 contained target 1. The second condition was HC, in which participants wrote the blue stimulus of simple Chinese characters on a writing pad and then responded to the same Chinese judgment target task. The third condition was drawing followed by Chinese recognition (DC), which asked participants to draw the priming stimulus (circle, square, triangle, diamond, rectangle, parallel lines, or wavy lines) on the writing pad first and then respond to the Chinese judgment target task. The fourth condition was drawing followed by English recognition (DE), in which participants drew the same priming stimulus as in the DC condition, followed by responding to the English target task.

Each participant participated in an EEG test with a total duration of 350 s. The data were collected in the EEG laboratory of the National Institute of Education Science, and all materials appeared in the center of the computer screen. Before the formal experiment, participants participated in a training activity

designed to familiarize them with the experimental procedures in all four conditions. See **Figure 1** for the flowchart of the presentation. To start, a fixation asterisk appeared on the screen for 200 ms; following the fixation, a blank black screen appeared for 300 ms. Then, there was a 2,000-ms learning phase. In all four conditions, the learning phase began with the stimulus in blue, followed by target 1 in red and then target 2 in white. In the handwriting condition, participants wrote the blue stimulus. In the viewing condition, participants spent the same length of time viewing the stimuli. After a blank black screen appeared for 1,000–1,500 ms (duration chosen at random), the red target 1 was shown to participants for 500 ms followed by a 500-ms blank black screen. Finally, target 2 appeared in white, and participants were instructed to press button “y” if target 2 included target 1 or button “n” if it did not. In a word, participants decided whether target 1 was included in target 2. When participants pressed the button, the screen disappeared; if no button was pressed, the screen remained for 3,500 ms. The program then advanced to the next trial. The EEG recording began upon the onset of the fixation, and continuous EEG recording proceeded, during which the responses to target 1 and target 2 were all marked in the EEG recording.

Event-Related Potential Data Acquisition and Preprocessing

Response time and accuracy were recorded during ERP data acquisition. ERP data were collected using NeuroScan’s ESI-64 system. Electrode position in this study approximated locations of the international 10–20 system. The study used the left mastoid as the reference electrode. The vertical electrooculogram (VEOG) was recorded using two electrodes placed above and below the midline of the right eye, and the recording electrodes of the

horizontal electrooculogram (HEOG) were placed beside the left and right eyes in horizontal alignment with the eyeball.

All electrodes were placed on the scalp using conductive paste to ensure that the impedance of each electrode was kept below 5 K Ω . The EEG data acquisition software was NEUROSCAN. The amplifier was SYNAMPS2, and AC continuous sampling was adopted. Scalp potentials were recorded with a sampling rate of 1,000 Hz, and the bandpass filter is 0.05~100 Hz.

Offline analysis of EEG data was performed using Curry 7.0. During the recording, the left mastoid was used; later, the data were referenced offline using a reference averaged across the left and right mastoids. First, a constant baseline correction was performed. Second, the data were digitally filtered with a 30-Hz lowpass. Then, the components related to eye movement were removed. In addition, amplitudes exceeding $\pm 100 \mu\text{V}$ were also excluded as artifacts. The continuous EEG data were segmented, with the duration of the segmentation starting 200 ms before the onset of target 1 and extending 800 ms after target 1. Finally, the ERP waves were superimposed and averaged, and the baseline correction was performed using the baseline of 200 ms before the stimulus.

Behavior and Event-Related Potential Data Analyses

For behavioral data, we conducted 4 (learning conditions: VC, HC, DC, and DE) \times 2 (children vs. adult as between-subject factor) repeated-measures analyses of variance (ANOVAs) on RT and ACC. For ERP data, according to prior literature (Maurer et al., 2008), the N170 component elicited by Chinese characters has generally been recorded on PO7 and PO8 electrodes, and a lateralization effect has been reported, with the left negative wave larger than the right negative wave (Rossion et al., 2007; Zhang et al., 2011). The stimulus-elicited peak and latency of the N170 at the PO7 and PO8 electrodes of each participant were extracted from the EEG data and analyzed by the statistical models by using SPSS 17.0.4. Here, 4 (learning conditions: VC, HC, DC, and DE) \times 2 (electrode position: left PO7 and right PO8) repeated-measures ANOVAs were performed to analyze the amplitude and latency of the N170 of both adults and children. After demonstrating a significant main effect of group and learning condition, as well as their interaction, we broke the analyses down into two groups (children and adults). To answer the first three research questions, we compared three pairs of learning conditions (VC vs. HC, VC vs. DC, DC vs. DE), and to answer the fourth research question regarding the laterality effect, we examined the hemispheric differences in the N170. To correct for multiple comparisons, a Bonferroni correction was applied because the data violated the assumption of sphericity (Bland and Altman, 1995; Chen et al., 2017). A significance level of 0.05 was used for all statistical analyses.

RESULTS

Behavioral Results

Because the adults and children were tested using the same materials and had all been trained on the procedures before

beginning the trials, behavioral differences between the adults and children can be attributed to their cognitive ability (Palmis et al., 2020). Therefore, behavioral data analysis did not focus on comparisons between adults and child but instead investigated the differences in behavioral performance in the four conditions between groups.

For behavioral data analyses, both ACC and RT for target 2 were collected. The aggregated means per subject per condition were submitted for ACC analyses. RTs were recorded from the onset of target 2 to the button press. Outliers were determined as those RTs located in the extreme 5% on either end of the Z-normalized distribution of RTs. This is equivalent to removing RTs above and below 1.65 SD of each individual participant mean RT. Overall, this resulted in 7.5% of trials being excluded as outliers, within the 5–10% recommended by Ratcliff (1993). **Table 1** shows the descriptive statistics of mean and SD of both ACC and RT for each of the four conditions. **Figures 2A,B** present violin plots summarizing the behavioral data for both children and adults.

Four repeated-measures ANOVAs were performed using a single factor (learning conditions: VC, HC, DC, and DE) by submitting RT and ACC for each condition across children and adult groups. The group (child vs. adult) factor was used as the between-participant factor. RT and ACC of children and adults demonstrated significant effects of learning condition. For RT, there was a significant effect of learning condition [$F(3,84) = 6.910, p = 0.003, \eta^2 = 0.198$] and condition \times group interaction [$F(3,84) = 4.297, p = 0.007, \eta^2 = 0.133$]. For ACC, there was a significant effect of learning condition [$F(3,84) = 64.539, p < 0.001, \eta^2 = 0.697$] and a significant condition \times group interaction [$F(3,84) = 29.951, p < 0.001, \eta^2 = 0.517$]. Therefore, three sets of *post hoc* analyses were carried out below in children and adults, respectively.

Comparing Handwriting vs. Viewing

Among children, the RT in HC ($M = 1,578 \text{ ms}$) was significantly shorter than that in VC ($1,734 \text{ ms}$) [$F(1,15) = 2.047, p < 0.001, \eta^2 = 0.68$], and the ACC rate in HC ($M = 0.98$) was significantly higher than that in VC ($M = 0.94$) [$F(1,15) = 334.657, p < 0.001$,

TABLE 1 | Mean and SD of both ACC and RTs in the four conditions.

Condition	RT			ACC		
	Adults	Children	Cohen's <i>d</i>	Adults	Children	Cohen's <i>d</i>
VC	779 (149)	1,734 (282)	1.71	0.88 (0.23)	0.94 (0.29)	1.44
HC	711 (137)	1,578 (261)	1.60	0.98 (0.17)	0.98 (0.27)	3.28
DC	739 (122)	1,628 (259)	1.63	0.90 (0.16)	0.94 (0.35)	0.36
DE	713 (137)	1,708 (301)	1.51	0.97 (0.19)	0.91 (0.33)	1.17

ACC, accuracy; RT, response time; VC, viewing Chinese; HC, handwriting Chinese; DC, drawing followed by Chinese recognition; DE, drawing followed by English recognition.

Standard deviation of each measure per condition presented in parentheses. We calculated Cohen's *d* by using the following formula: $[4\eta^2/(1-\eta^2)]^{1/2}$. Cohen's *d* < 0.2 indicates a small effect size, $0.2 < \text{Cohen's } d < 0.8$ indicates a medium effect size, and Cohen's *d* > 0.8 indicates a large effect size (Fritz et al., 2012).

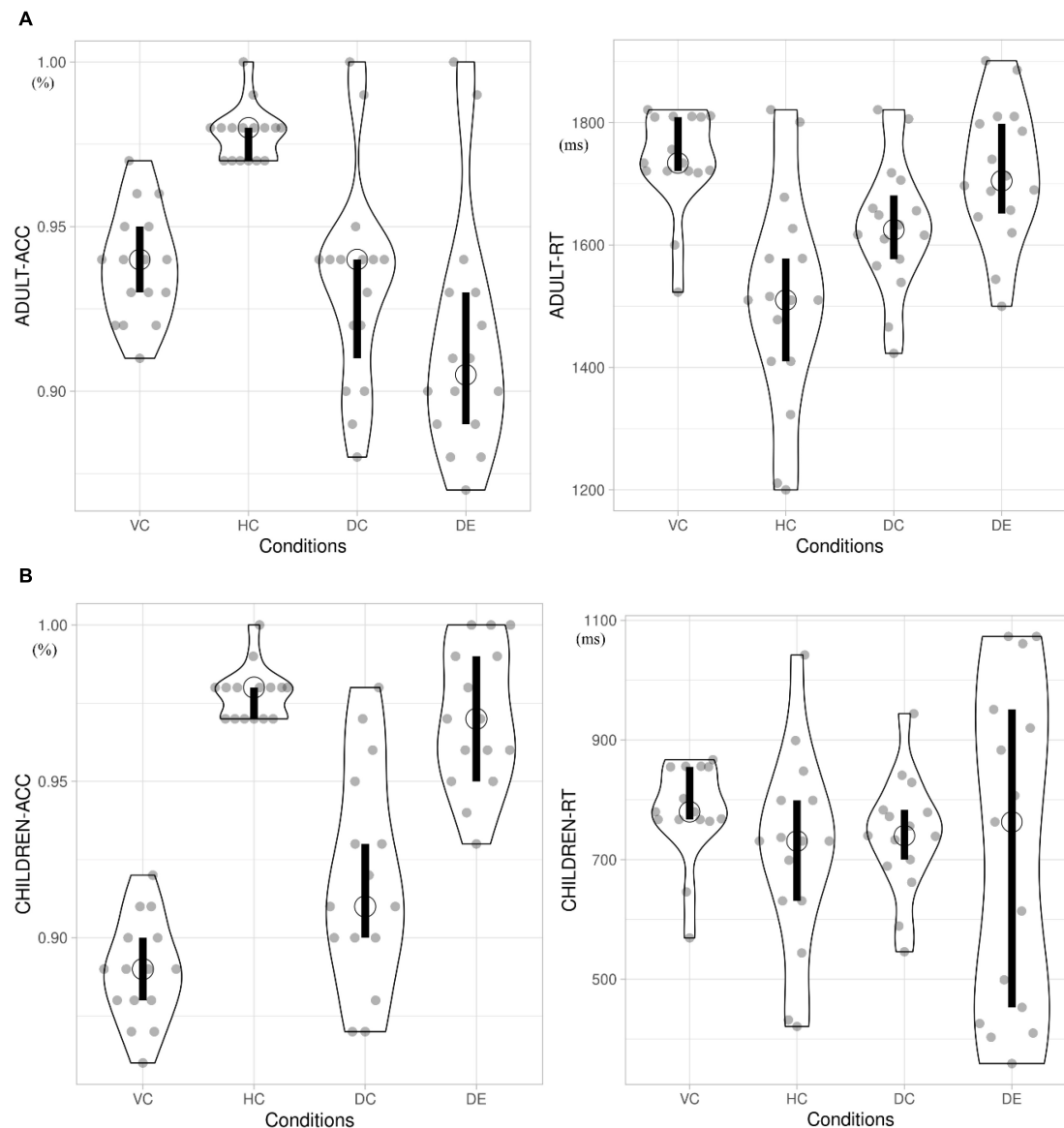


FIGURE 2 | (A) Behavioral data of adults. Open circle indicates the median in each condition. Vertical bar indicates the 95% confidence interval for each median determined by bootstrapping. ACC, accuracy; RT, response time; VC, viewing Chinese; HC, handwriting Chinese; DC, drawing followed by Chinese recognition; DE, drawing followed by English recognition. **(B)** Behavioral data of children. Open circle indicates the median in each condition. Vertical bar indicates the 95% confidence interval for each median determined by bootstrapping.

$\eta^2 = 0.923$]. For adults, the patterns were the same. Their RT in HC ($M = 711$ ms) was significantly shorter than that in VC ($M = 779$ ms) [$F(1,15) = 21.87, p < 0.001, \eta^2 = 0.422$], and ACC of HC ($M = 0.98$) was significantly higher than that of VC ($M = 0.88$) [$F(1,15) = 72.624, p < 0.001, \eta^2 = 0.708$].

Comparing Handwriting vs. Drawing Followed by Chinese Recognition

For children, the RT in HC ($M = 1,734$ ms) was significantly longer than that in DC ($M = 1,628$ ms) [$F(1,15) = 0.328, p < 0.001, \eta^2 = 0.012$], and the ACC in HC ($M = 0.98$) was significantly higher than that in DC ($M = 0.94$) [$F(1,15) = 41.502,$

$p < 0.001, \eta^2 = 0.597$]. For adults, there was a significantly longer RT of HC ($M = 779$ ms) compared with that of DC ($M = 739$ ms) [$F(1,15) = 5.278, p = 0.029, \eta^2 = 0.15$], and ACC in HC ($M = 0.98$) was significantly higher than that in DC ($M = 0.90$) [$F(1,15) = 30.198, p < 0.001, \eta^2 = 0.502$].

Comparing Drawing Followed by Chinese Recognition vs. Drawing Followed by English Recognition

For children, the RT of Chinese recognition in the DC condition ($M = 1,628$ ms) was not significantly different from that of English recognition in the DE condition ($M = 1,708$) [$F(1,15) = 0.132,$

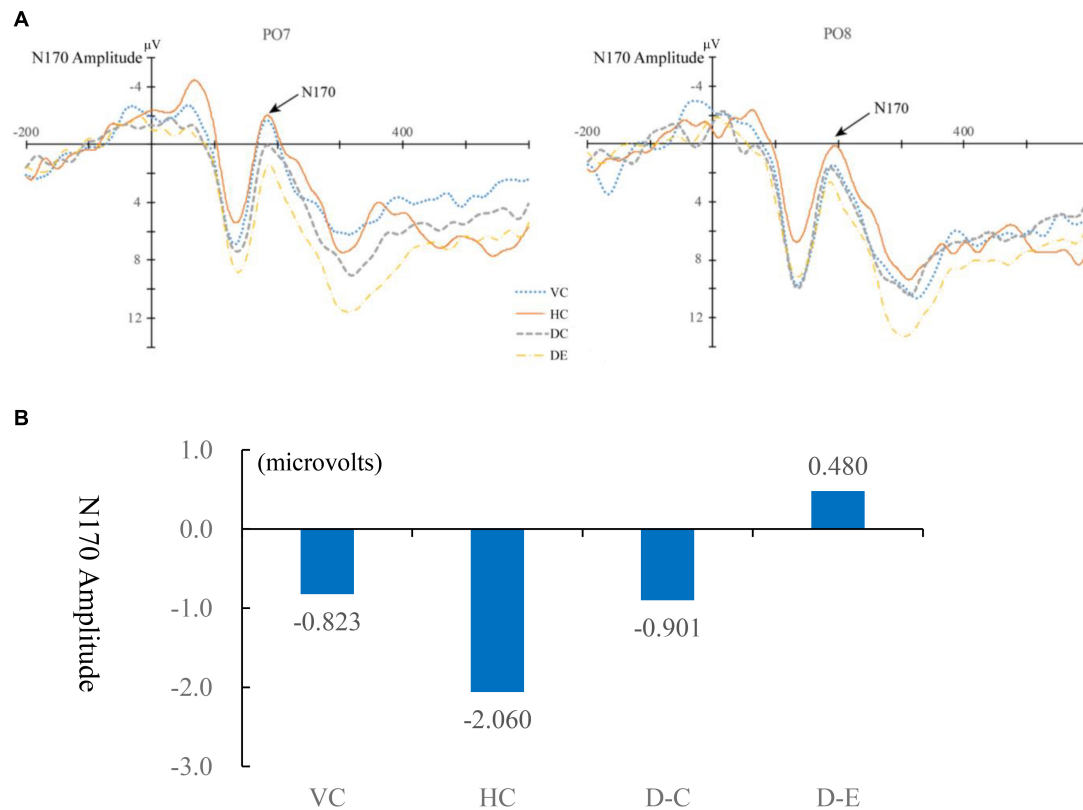


FIGURE 3 | (A) Event-related potential (ERP) waveforms of the N170 under four conditions for children for the left (PO7) and right (PO8) parietal leads. VC, viewing Chinese; HC, handwriting Chinese; DC, drawing followed by Chinese recognition; DE, drawing followed by English recognition. **(B)** Differences between the four conditions for children in the amplitude of N170.

$p = 0.719$, $\eta^2 = 0.005$], and the ACC of DC ($M = 0.98$) was higher than that of the DE condition ($M = 0.91$) [$F(1,15) = 23.083$, $p < 0.001$, $\eta^2 = 0.452$]. For adults, there was no difference between ACC [$F(1,15) = 2.047$, $p = 0.16$, $\eta^2 = 0.06$] and no difference in RT [$F(1,15) = 5.278$, $p = 0.08$, $\eta^2 = 0.15$] in the two drawing conditions.

Event-Related Potential Results

The original ERP waveforms that marked target 1 responses at PO7 and PO8 for children are shown in **Figure 3A** and for adults are shown in **Figure 4A**. A 4 (learning conditions: VC, HC, DC, and DE) \times 2 (hemisphere: left PO7 and right PO8) \times 2 (group: adult vs. children) repeated-measures ANOVA was carried out on the N170 amplitude. The results revealed a significant main effect of condition [$F(3,81) = 5.536$, $p = 0.002$, $\eta^2 = 0.165$], main effect of group [$F(1,28) = 5.344$, $p = 0.07$, $\eta^2 = 0.177$], and significant condition \times hemisphere \times group interaction [$F(3,81) = 0.954$, $p = 0.419$, $\eta^2 = 0.02$]. Moreover, we observed a significant two-way interaction of condition \times hemisphere [$F(3,81) = 6.858$, $p = 0.000$, $\eta^2 = 0.197$] and a significant two-way interaction of group \times hemisphere [$F(3,81) = 5.183$, $p < 0.001$, $\eta^2 = 0.152$], showing that there is a different pattern across hemispheres among the four conditions and between children and adults. Therefore, we broke down the N170 amplitude

analyses in a condition comparison within children and adult groups separately. **Table 2** shows the descriptive statistics of EEG data of all conditions. In addition, like previous studies (Maurer et al., 2008; Yum et al., 2014; Yum and Law, 2021), latency was analyzed, but the results were not significant, so we only report the EEG amplitude data results. Differences between the conditions are shown for children in **Figure 3B** and for adults in **Figure 4B**. **Table 3** shows the correlation matrix for behavioral and EEG data. **Figures 5A,B** present summaries of the N170 amplitude data for both children and adults.

Comparing Handwriting vs. Viewing

For children, there was a greater N170 amplitude during HC than that during VC [$F(1,15) = 0.72$, $p = 0.035$, $\eta^2 = 0.03$], showing that handwriting facilitates recognition of Chinese characters. For adults, the patterns were the same. The amplitude of the N170 was significantly greater for HC than for VC [$F(1,15) = 1.879$, $p = 0.029$, $\eta^2 = 0.059$].

Comparing Handwriting vs. Drawing Followed by Chinese Recognition

For both children and adults, there was no difference in N170 amplitude for HC and DC [$F(1,15) = 2.191$, $p > 0.05$, $\eta^2 = 0.068$ for adults; $F(1,15) = 0.473$, $p > 0.05$, $\eta^2 = 0.019$ for children].

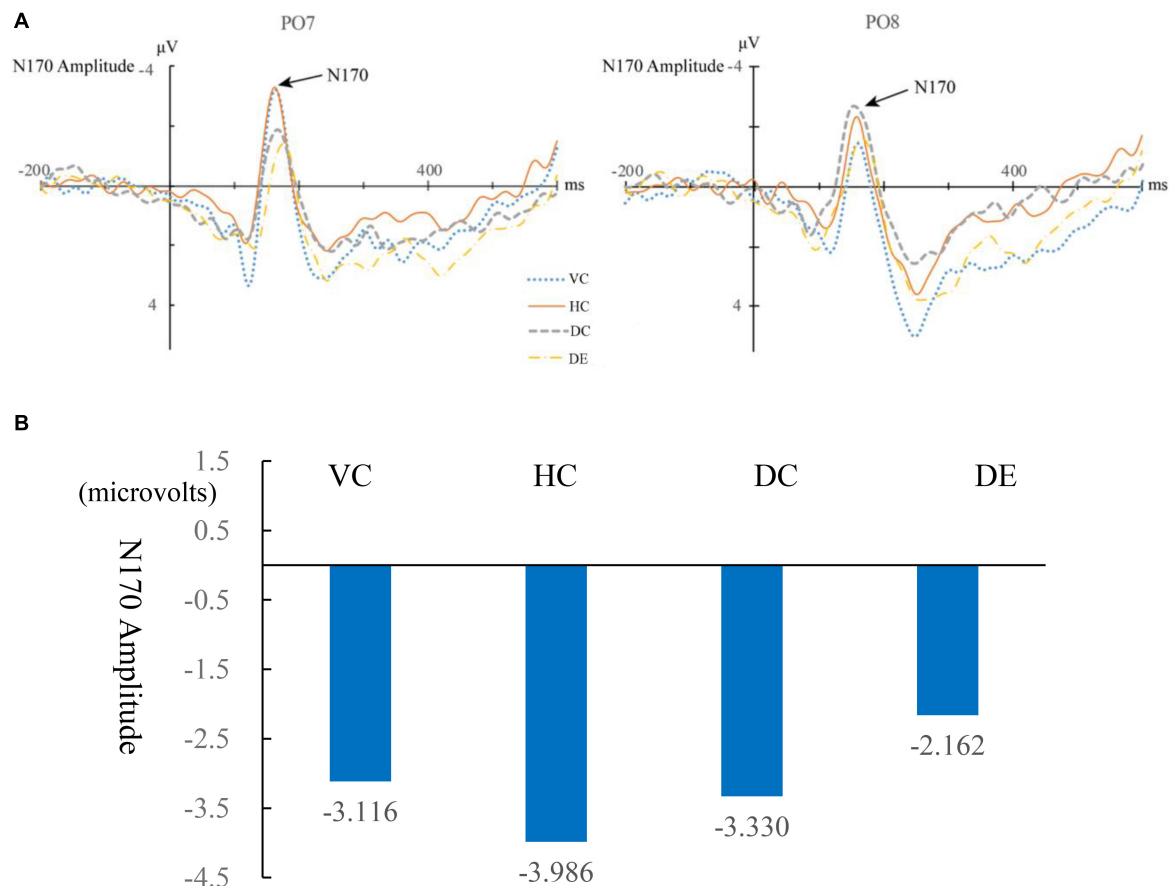


FIGURE 4 | (A) Event-related potential (ERP) waveforms of N170 under four conditions for adults for the left (PO7) and right (PO8) parietal leads. VC, viewing Chinese; HC, handwriting Chinese; DC, drawing followed by Chinese recognition; DE, drawing followed by English recognition. **(B)** Differences between the four conditions for adults in the amplitude of N170.

Comparing Drawing Followed by Chinese Recognition vs. Drawing Followed by English Recognition

For children, DC elicited a significantly larger N170 response than DE [$F(1,15) = 15.07$, $p = 0.02$, $\eta^2 = 0.53$]. For adults, the N170 amplitude was also greater for DC than DE [$F(1,15) = 0.527$, $p = 0.04$, $\eta^2 = 0.017$].

Laterality Effect

For adults, the peak value of N170 of the left hemisphere PO7 was significantly higher than that of the right hemisphere PO8 for HC [$F(1,16) = 7.794$, $p = 0.013$, $\eta^2 = 0.328$], VC [$F(1,16) = 9.208$, $p = 0.005$, $\eta^2 = 0.365$], but the laterality effects were not significant in the two drawing conditions [DC: $F(1,16) = 0.327$, $p = 0.572$, $\eta^2 = 0.011$; DE: $F(1,16) = 0.004$, $p = 0.948$, $\eta^2 = 1.461e^{-4}$]. For children, the four conditions showed no significant laterality [VC: $F(1,14) = 3.083$, $p = 0.091$, $\eta^2 = 0.110$; HC: $F(1,14) = 0.585$, $p = 0.452$, $\eta^2 = 0.023$; DC: $F(1,14) = 0.428$, $p = 0.519$, $\eta^2 = 0.016$; DE: $F(1,14) = 3.083$, $p = 0.091$, $\eta^2 = 0.110$]. **Figure 6** shows the lateralization of the N170 for the four conditions. Please see **Table 4** for a summary of the behavioral and N170 results.

DISCUSSION

We compared HC with VC characters and two other drawing conditions, i.e., drawing shapes followed by Chinese recognition (DC) and drawing shapes followed by English recognition (DE). There were four main findings. First, we revealed a facilitating effect, for both adults and children, of HC on behavior and the

TABLE 2 | Mean (SD) ERP magnitude at PO7 and PO8 for four conditions.

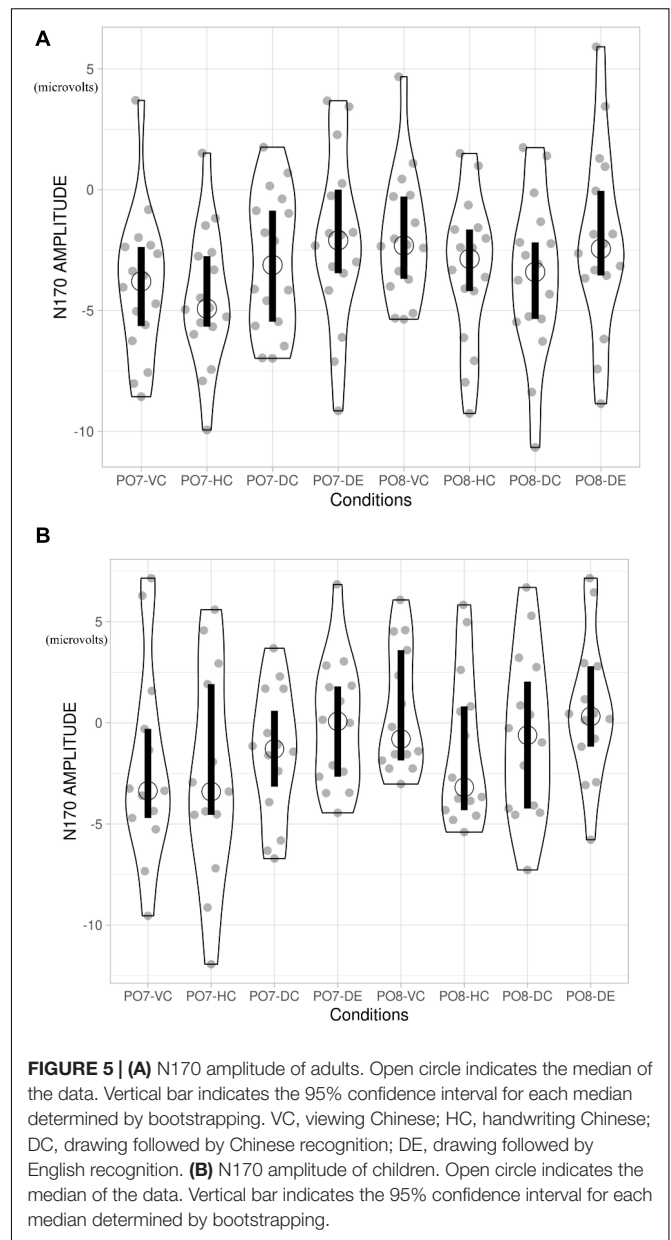
	Adults		Children	
	PO7	PO8	PO7	PO8
VC	-3.95 (2.96)	-1.98 (2.60)	-2.16 (4.67)	0.57 (2.96)
HC	-4.49 (2.74)	-3.37 (2.94)	-2.69 (5.06)	-1.35 (3.62)
DC	-2.99 (2.80)	-3.62 (3.22)	-1.54 (3.15)	-0.62 (3.96)
DE	-2.16 (3.46)	-2.07 (3.69)	-0.07 (3.08)	0.57 (3.38)

ERP, event-related potential; VC, viewing Chinese; HC, handwriting Chinese; DC, drawing followed by Chinese recognition; DE, drawing followed by English recognition.

Standard deviation of each measure per condition is presented in parentheses.

TABLE 3 | Correlational matrix between mean amplitude of P07 P08 and behavioral responses of RT and ACC.

	Adult								Children							
	RT				ACC				RT				ACC			
	VC	HC	DC	DE	VC	HC	DC	DE	VC	HC	DC	DE	VC	HC	DC	DE
P07	VC	0.263** (0.023)			-0.235* (0.008)				0.198 (0.177)				-0.210 (0.164)			
	HC		0.103 (0.254)		-0.302* (0.048)				0.348 (0.052)				-0.294 (0.183)			
	DC			0.122 (0.103)	0.059 (0.829)				0.151 (0.386)				-0.380** (0.007)			
	DE					0.087 (0.748)			-0.120 (0.135)				0.087 (0.767)			0.196 (0.501)
P08	VC	0.170 (0.158)			-0.270* (0.021)				-0.257 (0.376)				0.003 (0.993)			
	HC		0.208 (0.440)		-0.298 (0.127)				0.139 (0.116)				-0.048 (0.870)			0.182 (0.533)
	DC			0.176 (0.301)	0.188 (0.485)				0.003 (0.993)				0.163 (0.578)			0.081 (0.784)
	DE				-0.116 (0.668)				0.163 (0.578)				0.081 (0.784)			

* $p < 0.05$, ** $p < 0.01$; Significance levels are presented in parentheses.

N170 compared to VC. Second, we revealed a facilitating effect on ACC of HC on behavior measures compared to drawing shapes. Although we did not find neural effects, handwriting appears to enhance visual word recognition more than simply drawing shapes. Third, we found that drawing shapes appeared to have a larger effect on the N170 of Chinese characters compared to English words. Finally, we found a left lateralization of the effect of HC and VC, suggesting greater specialization in adults compared to children.

The facilitating effect on HC is represented by its comparison with VC, with shorter RTs and higher ACC in HC compared to VC. The peak of the N170 for HC was also significantly larger than that of VC. This ERP finding suggests that, in comparison to VC, HC enhanced the processing of Chinese characters for

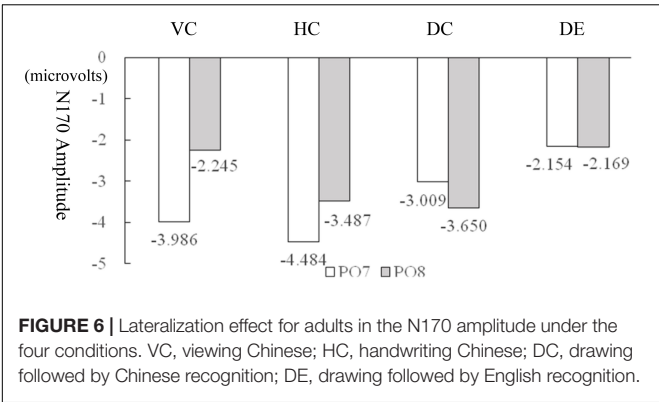


FIGURE 6 | Lateralization effect for adults in the N170 amplitude under the four conditions. VC, viewing Chinese; HC, handwriting Chinese; DC, drawing followed by Chinese recognition; DE, drawing followed by English recognition.

both adults and children. The finding that a stronger N170 was triggered by the HC than the VC condition suggests that the N170 indicates enhanced orthographic word recognition. This finding is consistent with the results of Liu and Perfetti (2003), who found this pattern for Chinese–English bilinguals, and with a series of handwriting training studies (Guan et al., 2011, 2015, 2020; Guan and Fraundorf, 2020). Handwriting training appears to enhance familiarity with the orthographic representation of the word. This finding is also consistent with a study with artificial orthographies by Yoncheva et al. (2010), who found that the unit size acquired through training influences N170 response to visual words, which was greater when training was based on the small unit size (i.e., grapheme compared to whole word). For both children and adults in our study, handwriting training drew more attention to the small units within the word form. The judgment task asked them to decide whether a simpler character was embedded in the more complicated whole character. Paying attention to the local features may enhance the early processing of Chinese characters, thus affecting the N170.

Handwriting practice likely increases motor–sensory integration to facilitate visual recognition by focusing on the detailed visual–orthographic components of stroke composition (Guan et al., 2011). Guan et al. (2015) found that the improvement of handwriting quality predicted gains in reading comprehension when previous knowledge was controlled for. Handwriting has a sensory–motor source for native language, forming a mental model accompanied by a new neural motor memory (Shadmehr and Holcomb, 1997). Sensory–motor training facilitates language cognition (Guan and Wang, 2017). That is, people who can better understand the visual–motor coupling in this language are usually those who more effectively learn the visual–orthographic representation of the written language.

The higher ACC level for HC than DC revealed that HC characters led to better performance than drawing followed by Chinese recognition, suggesting that handwriting helps to coordinate the brain, eyes, and fingers to establish a subtle representation for sub-lexical word forms (Guan et al., 2011). Handwriting may accelerate the perception of Chinese characters for both adults and children (Guan et al., 2015). However, the reaction times for DC were faster than those for HC for both adults and children, and the EEG results for HC and DC were not

TABLE 4 | Summary table of behavioral and EEG results.

		HC vs. VC	HC vs. DC	DC vs. DE	
Adults	ACC	> (1.46)	> (1.23)	> (1.16)	
	RT	< (1.13)	> (0.67)	ns	
	N170	> (0.16)	ns	> (0.22)	
		HC	VC	DC	DE
	Laterality	L > R (0.99)	L > R (1.05)	ns	ns
Children	ACC	> (1.66)	> (1.34)	ns	
	RT	< (0.61)	> (0.19)	ns	
	N170	> (0.09)	ns	> (1.26)	
		HC	VC	DC	DE
	Laterality	ns	ns	ns	ns

Effect sizes represented by Cohen's *d* for the group comparison are reported in the parentheses. We calculated Cohen's *d* using the following formula: $[4\eta^2/(1-\eta^2)]^{1/2}$. Cohen's *d* < 0.2 indicates a small effect size, 0.2 < Cohen's *d* < 0.8 indicates a medium effect size, and Cohen's *d* > 0.8 indicates a large effect size (Fritz et al., 2012).

EEG, electroencephalogram; VC, viewing Chinese; HC, handwriting Chinese; DC, drawing followed by Chinese recognition; DE, drawing followed by English recognition; ACC, rates for the binary decision; RT, response time; L, left hemisphere; R, right hemisphere.

significantly different. These mixed results suggest that the N170 may be influenced by both handwriting and drawing.

The different performance in DE and DC may possibly reflect differences in the ways adults and children process Chinese and English. Foremost, our results comparing between DC and DE may just reflect the language difference effect itself. Processing of Chinese may involve a category-specific form of processing. Indeed, a larger N170 has been observed for Chinese characters relative to English, along with a more left-lateralized N170 for Chinese characters for English–Chinese bilinguals compared to English-only participants (Wong et al., 2005). Therefore, the processing of Chinese may, like faces, involve “special” processing in the brain, although the hemispheric lateralization of the N170 to such stimuli is still unclear.

Meanwhile, there was an enhancement of the N170 in drawing followed by Chinese recognition (DC) compared with drawing followed by English recognition (DE), probably reflecting a native language effect. Most children in China only begin to learn English in the third grade. In our study, Chinese was the native language for all participants, and therefore, they were much more familiar with Chinese characters than English letters. Our finding that native Chinese-speaking adults and children displayed a greater N170 effect on Chinese than their second language (English) is consistent with findings of Liu and Perfetti (2003) that the N170 perceptual effect of a native language was greater than that of a second language. Research has shown that the N170 indexes visual–orthographic processing. Orthographic stimuli (such as words, pseudo-words, and consonant strings) produced greater N170 effects than non-orthographic stimuli (such as symbols) (Bentin et al., 1999; Pylkkanen and Marantz, 2003; Simon et al., 2004). Chinese adults and children are much more familiar with Chinese than English, which may have produced a larger N170 component.

Adults showed a lateralization of the N170 effect in the HC and VC conditions, but the children did not. Adults have developed much experience with written language; therefore, they show N170 lateralization during the viewing and handwriting conditions. People are not born with N170 lateralization nor does it exist in early cognition in children. Rather, it is the result of humans' experience with written language in their later years. This pattern of results is in line with the existing literature that has found a left-lateralized effect of the N170 for Chinese characters (Maurer et al., 2008). Previous studies have reported an enhanced N170 for words in syllabic writing systems compared to control stimuli but did not explicitly test left lateralization (Kim et al., 2004; Shirahama et al., 2004). In addition, left lateralization has been shown to be characteristic of visual expertise for words written in alphabetic scripts (Bentin et al., 1999; Rossion et al., 2003; Maurer et al., 2005b). The current results suggest that similar processes underlie the left-lateralized N170 in logographic writing systems and writing systems that associate characters with larger phonological units, such as syllables.

Remarkably, Cao et al. (2011) tested all four age groups (7-, 9-, and 11-year-olds, as well as college students); even the youngest group showed a left-lateralized N170 response for Chinese characters, suggesting that a relatively specialized mechanism for processing Chinese characters is already emergent by as early as 7 years of age. However, our results showed that adults demonstrated laterality, while children ($M_{\text{age}} = 9.5$ years) did not. Visual form familiarity serves as an important driver for the increased and left-lateralized N170 response among adults. Xue et al. (2019) found an increased and left-lateralized N170 response for regular characters compared to cursive characters that were less familiar. It is possible that the amount of training was not sufficient for increasing the familiarity of the visual characters for the children in our study.

Our study is not without limitation. Because we used the same stimuli across groups, the difficulty level of our stimuli was not the same in children and adults. Future research should consider the varying difficulty levels across ages. In addition, because the participants only engaged in handwriting or drawing for a few seconds, the modest effects might be due to the shorter duration. Longer exposure to the learning conditions might lead to greater effect sizes. Children might benefit from longer handwriting experiences in those conditions. In addition, handwriting curved letters in comparison to the straight-line letters/characters might have different effects on the brain's visual-form areas (Ose Askvik et al., 2020). Finally, more fine-grained examination of the EEG before 170-ms post stimulus onset might also be considered (Woodman, 2010), as this might reveal an effect of handwriting on sensory processing (Pratt, 2011), word recognition (Hillyard et al., 1998), or visual discrimination (Vogel and Luck, 2000).

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CONCLUSION

We found that HC produced a larger N170 and better performance than VC and better performance than drawing shapes for both children and adults. The key mechanism under these effects may be visual-motor integration. The interaction between visual and motor areas may enhance orthographic representations. The left lateralization of the N170 effect was seen in adults and not children, suggesting that greater familiarity with characters and more practice with handwriting are necessary to improve the quality of the orthographic representations in children. Future studies should further explore different methods to facilitate orthographic perception through handwriting.

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 Science and Technology Beijing (USTB).

AUTHOR CONTRIBUTIONS

CG and WM designed the study, collected and analyzed the data, wrote the manuscript. ES and JB provided the comments and proofed the manuscript. All authors contributed to the article and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fpsyg.2021.628160/full#supplementary-material>

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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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Visual Neuropsychology in Development: Anatomico-Functional Brain Mechanisms of Action/Perception Binding in Health and Disease

Silvio Ionta*

Sensory-Motor Lab (SeMoLa), Department of Ophthalmology-University of Lausanne, Jules Gonin Eye Hospital-Fondation Asile des Aveugles, Lausanne, Switzerland

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Edited by:

Cosimo Urgesi,
University of Udine, Italy

Reviewed by:

Shahar Arzy,
Hadassah Medical Center, Israel
Satoshi Nobusako,
Kio University, Japan

*Correspondence:

Silvio Ionta
ionta.silvio@gmail.com

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Vision is the main entrance for environmental input to the human brain. Even if vision is our most used sensory modality, its importance is not limited to environmental exploration. Rather it has strong links to motor competences, further extending to cognitive and social aspects of human life. These multifaceted relationships are particularly important in developmental age and become dramatically evident in presence of complex deficits originating from visual aberrancies. The present review summarizes the available neuropsychological evidence on the development of visual competences, with a particular focus on the associated visuo-motor integration skills in health and disease. With the aim of supporting future research and interventional settings, the goal of the present review is to constitute a solid base to help the translation of neuropsychological hypotheses into straightforward empirical investigations and rehabilitation/training protocols. This approach will further increase the impact, ameliorate the acceptance, and ease the use and implementation of lab-derived intervention protocols in real-life situations.

Keywords: vision, movement, brain, child, sensorimotor

NEURAL CORRELATES OF VISION

Visual perception permeates our life, not only for merely gathering information about the environment, but also for having important influence on our motor skills. Revealing the neural mechanisms of the multifaceted relationships between vision and other domains of human life, visual neuropsychology goes beyond the traditional consideration of vision as a passive function, and rather highlights how visual competences can impact typical and atypical development at a more systemic, dynamic, and integrated level. The neurobiological machinery that brings from light to vision starts in the eyes, where the photoreceptors of the retina are able to selectively respond to the photons of light (entered through the cornea and projected to the retina) and “translate” them into neural signals. These signals are transported by the optic nerves to subcortical structures (the lateral geniculate and pulvinar nuclei of the thalamus) which relay signals mainly to the visual cortex, in the posterior part of the brain, but also to the superior colliculus in the midbrain (Shipp, 2004). The occipital lobe is further organized in several sub-regional areas, including the striate primary visual cortex (V1) and a series of interconnected, extra-striate, and progressively more

specialized areas for higher-level processing of visual input (Figure 1). Thus, while V1 is sensitive to basic features of the visual input, such as line orientation, motion direction, and depth perception, the secondary visual cortex (V2) receives fibers from V1, projects to the third visual cortex (V3), and is already able to perform figure/background distinctions (Qiu and Von Der Heydt, 2005; Maruko et al., 2008), to process illusory contours (Von Der Heydt et al., 1984; Anzai et al., 2007), and to build binocular disparity (Von Der Heydt et al., 2000). V3 projects to areas out of the occipital lobe, including the posterior parietal cortex (Stepniewska et al., 2016) and the inferior temporal cortex (Ponce et al., 2017), and is sensitive to global motion (Braddick et al., 2001), covering larger portions of the visual field with respect to V1 (Lui et al., 2006). The fourth visual cortex (V4) is tightly connected to V1 and V2 (Liu et al., 2020) and projects mainly to the inferior temporal cortex (Bohon et al., 2016). It is involved in color perception, object recognition, and is sensitive to top-down attentional modulation (Roe et al., 2012). The fifth visual cortex (V5) receives input from V1, V2, V3, as well as from the thalamus (Ungerleider and Desimone, 1986; Felleman and Van Essen, 1991; Sincich et al., 2004; Warner et al., 2010) and projects to the superior temporal gyrus (Handa et al., 2017; Handa and Mikami, 2018), the frontal eye fields (Machner et al., 2010) and lateral intraparietal cortex (De Azevedo Neto and Amaro Junior, 2018). Some fibers reach V5 directly from the thalamus, bypassing V1 (Warner et al., 2012). Encoding speed and direction of visual input (Dubner and Zeki, 1971; Maunsell and Van Essen, 1983), V5 is mostly important for motion perception and smooth guidance of eye movements (Dursteler et al., 1987) as well for “building” a continuous perception of moving targets and scenes instead of a “crystallized” vision of distinct frames (Hess et al., 1989; Baker et al., 1991). The sixth visual cortex (V6) is located medially and connected to parietal and pre/post-central regions (Shipp et al., 1998; Galletti et al., 2001; Luppino et al., 2005; Smith et al., 2018; Serra et al., 2019) of the brain is responsible for “subtracting out” the visual input related to self-motion from the rest of the visual perception (Pitzalis et al., 2013), as well as for visually guiding movements (Pitzalis et al., 2015).

Traditional neuropsychological models of visual perception indicate that the several interconnections among the visual regions of the brain can be broadly classified according to two functionally different main streams: the well-known ventral and dorsal streams (Tong, 2003). The “what” *ventral* stream would pass signals from V1, V2, V3, V4, up to the inferior temporal cortex and would be implied mainly in object recognition. Conversely, the “where” *dorsal* stream would comprise connections between V1, V2, V3, superior/medial temporal sulcus, and parietal cortex and would be particularly important for neurally encoding the visuo-spatial and motion-related aspects of visual input (Hickok and Poeppel, 2004; Almeida et al., 2010; De Haan and Cowey, 2011; Goodale, 2013). Lesions in the ventral stream produce recognition deficits such as prosopagnosia (the impossibility to recognize faces) (Mayer and Rossion, 2007). Lesions in the dorsal stream determine visuo-motor deficits, such as optic ataxia (impaired visuo-motor coordination, e.g., impossibility to reach objects despite

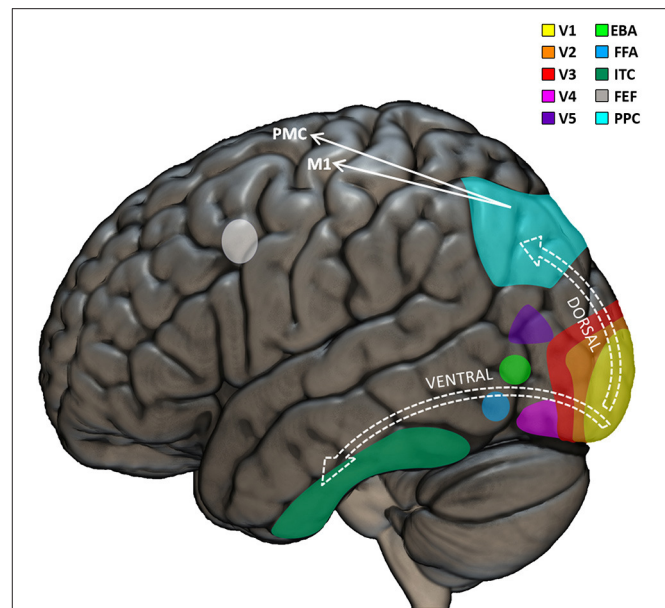


FIGURE 1 | Visual neuropsychological model. Graphical representation of the main cortical regions involved in visual perception and visuo-motor coordination. The visual input is first processed by the primary visual cortex (V1). Further processing is performed by the extrastriate visual regions (V2, V3, V4, and V5) which triggers the recruitment of the dorsal or ventral stream as a function of whether or not the visual input needs to be used to perceive or move in the environment, respectively. EBA, extrastriate body area; FFA, fusiform face area; ITC, inferior temporal cortex; FEF, frontal eye field; PPC, posterior parietal cortex; M1, primary motor cortex; PMC, premotor cortex.

preserved visual and motor skills separately) (Himmelbach et al., 2009). This sharp dichotomy between ventral and dorsal streams has been progressively smoothened (Rossetti et al., 2017), including the identification of bidirectional interactions between the streams (Greulich et al., 2020), especially in the context of adaptive behavior (Goodale et al., 2005) and visuo-motor skills (Van Polanen and Davare, 2015). Interestingly, different visuo-motor sub-pathways have been identified in the dorsal stream: the dorso-dorsal stream would be recruited for online action control; the ventro-dorsal stream would have been involved in higher-level cognitive functions including action understanding (Rizzolatti and Matelli, 2003). Altogether, it seems clear that the fine precision of our visual skills and their importance in action-related mechanisms are reflected in the high complexity of the involved neural architecture.

NEURO-BEHAVIORAL DEVELOPMENTS OF VISUAL SKILLS

Taking into consideration the temporal aspects of the development of the visual streams, it appears that the pace at which the ventral and the dorsal streams grow would be different already in pre-born age, indicating that the ventral stream would mature more quickly than the dorsal stream (Tadros et al., 2015). Indeed, already at birth the ability of

newborns to notice that a visual event occurs, even if the classification of “what” that even is still need further cortical maturation, has been considered an example of the importance of subcortico-cortical visual functions (Bronson, 1974). In addition, the fact that newborns are particularly attracted by face-like visual stimuli (Simion et al., 2011) and, especially, by familiar faces (Bushneil et al., 1989), suggests the functioning of thalamo-V5 connections which would bypass V1. Then, during the first 6 months, more specific functions associated with neural activity in V1 progressively emerge in an ordered sequence. The first functions are sensitivity to orientation, followed by the ability to perceive directional motion, and finally binocular interactions, e.g., for depth perception (Braddick and Atkinson, 2011).

Basic Functions

Visual Acuity

Visual acuity refers to our ability to perceive fine visual details. To reach adult levels, visual acuity rapidly develops during the first months of life and keeps improving up to 3 or 4 years (Norcia and Tyler, 1985; Banks and Dannemiller, 1987). Together with visual acuity, at birth also contrast sensitivity (the ability to discriminate light and dark) is below adult levels and progressively improves during the first months (Banks and Salapatek, 1978). From an ecological perspective, newborns don't need to perceive small details of far objects, but rather they need to recognize persons relevant for them (e.g., parents). Thus, newborns' relatively low visual acuity anyways allows them to efficiently interact with the environment relevant at their scale, even if their visual performance would be rated as “blindness” according to adult scales. Their low visual acuity might derive from the immaturity of foveal photoreceptors, retina, and eye-brain pathways, which quickly develop in early life, including denser concentration and better sensitivity of photoreceptors (Yuodelis and Hendrickson, 1986) and larger neural sprouting between the eyes and the brain (Braddick and Atkinson, 2011).

Vernier Acuity

The simple visual acuity provides the means to perceive small details, but it does not take into account the spatial relationship between such details. This capacity is defined “vernier acuity” and refers to the ability to perceive spatial incongruence, e.g., misalignment, with a resolution even higher than simple visual acuity. The behavioral aspects of vernier acuity have been long known, since the seminal work by Ewald Hering at the end of the nineteenth century (Strasburger et al., 2018). As it goes beyond the physical features of the eye, the vernier acuity is one of the examples of the importance of cortical dynamics in supporting visual skills (Manny, 1988; Skoczenski and Norcia, 1999). Indeed it progressively improves faster than the simple visual acuity (Zanker et al., 1992; Brown, 1997), in parallel with the maturation of the cortico-subcortical networks responsible for the integration of spatial relationships between different objects and their parts, and it reaches maturity much later than simple acuity (Skoczenski and Norcia, 2002). Interestingly, Braille reading triggers a progressive improvement of vernier

acuity on tactile perception (Loomis, 1979), suggesting the influence of neuroplastic changes driven by specific habits.

Accommodation

Both visual acuity and vernier acuity depend on accommodation, the possibility to adapt the focus to the distance of the object by contracting or relaxing the muscles of the eye lens. Accommodation at birth is limited to objects located within a range of 40–50 cm (Horwood and Riddell, 2008), with a possibly parallel ongoing attentional limitation (Downey et al., 2017). At the neural level, accommodation is associated with brain activity in the visual cortex (Mirzajani et al., 2017) and relies on an extended cortico-cerebellar network, including links of the visual cortex with cerebellar hemispheres/vermis and temporal cortex (Richter et al., 2000), as well as with precentral and frontal regions (Lv et al., 2020).

Color

All visual functions would not reflect the real world if they would not comprise information about colors. Color perception is strongly based on the early activity of cone receptors in the eyes from the first months of life (Brown, 1990). It has roots in the development of cortical functions starting from V2 and V3 (Ting Siok et al., 2009) to allow the proper use of color information including, for example, high-level functions such as emotion (Yoto et al., 2007) or aesthetics (Maglione et al., 2017).

Integrative Functions

Contours and Motion

One the most important information about the environment refers to the contours of an object: where an object ends and another one or the background start. This type of information is not present in the raw visual input, but it is rather built by neural responses in V1 (Hubel and Wiesel, 1977). Newborns start to discriminate orientation and therefore contours within the first weeks (Slater et al., 1988), even before visual event related potentials (VERPs; the stereotyped brain response to a standardized basic visual input) associated with contours can be recorded from their brains, between 3 and 8 weeks from birth (Braddick et al., 1986). Considering the importance of sensitivity to contours in development (Candy et al., 2001), VERPs can be fundamental measures for early detection of at-risk populations (Atkinson et al., 2008). Another fundamental information to efficiently understand the environment is the ability to distinguish static and moving objects. Like contours, motion sensitivity is the result of a cortical construction based on the neural responses in the visual cortices. Usually, newborns start to perceive motion a bit earlier than 10 weeks (Braddick et al., 2003), when their VERPs start to be detectable (Wattam-Bell, 1991). The anatomical maturation necessary to discriminate contours and motion occurs in the first months of life, depending on neural sprouting and synaptic establishment (Huttenlocher et al., 1982). The functional development of sensitivity to orientation and motion (plus binocularity) occurs in sequential order and based on neurally distinct pathways. For example, even if newborns show some degrees of motion sensitivity in relatively

early stages, this ability relies mostly on sub-cortical structures (Morrone et al., 1999).

Global Motion

The perception of motion alone does not account for the complexity of the environment, where different objects can move at a different pace and/or according to different spatio-temporal patterns. The capacity to perceive such a complexity seems to appear already between the first 2–6 months of life (Kellman and Spelke, 1983; Arterberry and Yonas, 2000; Johnson and Mason, 2002), with a specific ability for the recognition of human action-like motion, or biological motion (Booth et al., 2002). Such a relatively late development together with the need to convey information from larger areas of the visual field suggest that global motion skills rely on a neural architecture starting in V1 (Robertson et al., 2014) and further extending to a broader brain network (Koyama et al., 2005), likely encompassing V2 and V3 (Furlan and Smith, 2016), as well as V5 (Giaschi et al., 2007) and possibly also more frontal motor regions (Saygin et al., 2004; Wuerger et al., 2012). Already in about 5-year-old children, the perception of global motion is independent from simple visual acuity (Chakraborty et al., 2015) and is strictly related to visuo-motor skills (Chakraborty et al., 2017). In addition, sensitivity to global motion is neurally dissociable from global form perception (Vachon et al., 2009), possibly being linked to interregional neural connections (Pavlova et al., 2005). A direct way to assess global motion skills is the so-called “motion coherence sensitivity” test (Newsome and Pare, 1988), which evaluates the ability to recognize target motion patterns within a background of differently moving dots. During the first months of life, motion coherence sensitivity increases progressively (Mason et al., 2003), the associated VERPs show specificity for global motion (Wattam-Bell et al., 2010), and V5 is selectively activated by global motion in connection with other motion-related areas (Biagi et al., 2015).

Static Forms vs. Global Motion

There seem to be a clear segregation between sensitivity to global motion and static forms, as shown by the earlier readiness of VERPs associated to global motion than to static forms (Wattam-Bell et al., 2010), the larger variability of thresholds for global motion than for static forms (Braddick et al., 2016), and qualitative difference between global motion and static forms skills ranging up to adolescence (Meier and Giaschi, 2014). At the neural level, while sensitivity to global motion seems to recruit mainly the dorsal stream, the perception of static forms is mainly bound to the ventral stream. Accordingly, only the development of global motion perception (not static forms) correlates with anatomo-functional growth of neural connections between the parietal and frontal lobes, beyond the solely visual cortex (Braddick et al., 2016, 2017). This suggests that global motion perception is a higher-order function recruiting also sensorimotor integration mechanisms. Such a conclusion is further supported by the observation that the performance in global motion (not static forms) correlates with visuo-motor skills already in developmental age (Braddick et al., 2016) and that aberrancies in its parieto-frontal neural architecture might

be at the origin of the so-called “dorsal stream vulnerability” in developmental deficits (Spencer et al., 2000).

3D and Depth Perception

The ability to merge and coordinate information from the two perspectives of each eye provides one of the means to perceive three-dimensionality and is a peculiar cortical function, not happening in earlier levels of the visual input's processing. In children, binocularity emerges at about 3 months (Thorn et al., 1994), depending on ocular convergence (Downey et al., 2017), cortical maturation (Elberger and Smith, 1985), neural plasticity (Chalupa, 2004), specific neurotransmitters (Kameyama et al., 2010; Krahe and Medina, 2010), and cortico-cortical interactions, both in humans (Jurcoane et al., 2007) and other mammals (Dehmel and Lowel, 2014). The failure of one or more of these cortical mechanisms can contribute to the creation of the conditions for developing binocularity-related functional deficits, such as strabismus (Berman and Murphy, 1981; Freeman et al., 1982; Di Stefano and Gargini, 2002). Together with binocularity, depth perception relies on a number of visual abilities, including shape/shade segregation, sensitivity to differential texture density, interposition of near/far surfaces, all of which start to be present between 4 and 7 months (Yonas et al., 2002), and it keeps progressing in parallel with the development of fine visuo-motor skills both in health (Braddick and Atkinson, 2013) and disease (Grant et al., 2014). A specific function tightly linked to depth perception is the ability to identify an object with respect to its background. Such a figure-ground discrimination can be based, for example, on the sensitivity to different textures between the object and the background which starts to emerge in the first month of child's life (Brooks and Clair, 1971; Wattam-Bell, 1992), keeps evolving up to adulthood (Anderson et al., 2016), and is sensitive to age-related ocular diseases like macular degeneration (Tran et al., 2011). A related effect refers to the perception of the so-called “illusory contours,” proper visual illusions inducing the illusory perception of edges without physical borders, as shown by the pioneering work by Gaetano Kanisza and his famous illusory triangle (Kanisza, 1955). Children start to be sensitive to the Kanisza triangle in the first 3–5 months of life (Kavšek, 2002; Otsuka and Yamaguchi, 2003), with increasing sensitivity up to adolescence (Bondarenko et al., 2010), possibly in parallel with improved cortico-cortical interactions (Fyfytche and Zeki, 1996), increased intracortical dynamics in V2 and in V2-V5 exchanges (Grossberg, 2014), and exchanges between the different compartments of the visual cortex (Weigelt, 2007). Altogether, bidirectional interactions seem to be in place between children's improvements in perceiving basic visual features and their developments in higher-level functions beyond the mere visual perception. Such interactions typically start in the first month of life (Granrud, 2006) and keep evolving up to about 10 years (Nardini et al., 2010; Dekker et al., 2015).

Face Perception

The human face is probably the most salient visual stimulus in our life. The evolutionary and adaptive importance of the recognizing, understanding, and interpreting human faces is

demonstrated by the existence of a region in the extrastriate cortex of the human brain, specifically dedicated to processing face-related visual input: the fusiform face area or FFA (Kanwisher et al., 1997). Behavioral evidence shows that sensitivity to faces is one of the first visual abilities in newborns (Goren et al., 1975; Ferrari et al., 1986), is based on contrast polarity (Rosa Salva et al., 2012), can be functional even with a relatively low resolution at short distances (Von Hofsten et al., 2014), and can be shaped by life experience (Cobbett and Snelgrove-Clarke, 2016). At the neural level, the early appearance of the strong bias toward faces suggests that it is based on subcortical mechanisms aiding the newborn to fixate a face which in turn would favor a frequent exposure to faces and the associated development of selective cortical processing. This idea is supported by the supposed presence of visual pathways which would allow face perception by directly connecting the thalamus and the amygdala to FFA, bypassing V1 in both the human (Morris et al., 2001) and the primate brain (Bourne and Morrone, 2017). Despite cortical electrophysiology suggests that the FFA is sensitive to observation of faces already at 4 months (De Heering and Rossion, 2015), face-related neural response (De Haan et al., 2003) and cortical specialization (Peelen et al., 2009; Deen et al., 2017) seem less pronounced in children than adults.

Body Perception

Together with face perception, also the visual perception of the human body plays a crucial role in daily life. Similarly to the fusiform face area, and in obvious anatomical closeness, the extrastriate body area (EBA) is functionally-defined brain regions, specifically sensitive to the observation of the human body (Downing et al., 2001) and part of the lateral occipitotemporal cortex possibly overlapping with V5 (Ferri et al., 2013). The inhibition (Urgesi et al., 2007; Candidi et al., 2008) or lesion (Peelen and Downing, 2007) of EBA support its causal implication in selectively respond to the observation of human bodies. Not only is EBA important for body-related visual processing, but also it is involved in higher-level visual cognition related to the human body, including identity attribution (Myers and Sowden, 2008), emotional resonance (Ionta et al., 2020), and mental imagery (Arzy et al., 2006; Costantini et al., 2011; Perruchoud et al., 2016). Even if the development of EBA in the life span remains largely unexplored, recent evidence suggests that the development of the body-specific responsiveness of EBA can take several years. Indeed neuro-functional differences of EBA can be noticeable between 6 and 8 vs. 9–12 year-old children (Walbrin et al., 2020), and the development of EBA can be affected by neurological disorders in early age (Okamoto et al., 2017).

Visuo-Motor Interactions

Moving is one of the most direct and evolutionary relevant reason to have vision. Thanks to movements we can preserve our body, and therefore our life, for example by escaping dangers and reaching targets. Even if these functions can be technically possible also without vision, in typical conditions human beings are historically hardwired to vision. Therefore, it is not surprising that a large part of visual functions is subsequently used to control

movements, as suggested by tight visuo-motor resonance already in early age (Lepage and Théoret, 2007). The neural architecture for such visuo-motor couplings would be present already at birth (Meltzoff and Moore, 1977) and it would be promoted by the repeated exposure to sensorimotor events (Cook et al., 2014), being its development further depending on experience (Simpson et al., 2014).

Eye and Head Movements

The first visuo-motor interactions in the newborns comprise eye and head movements, followed by postural adjustments, manual exploration, and locomotion (Adolph and Franchak, 2017). Eye movements constitute a fundamental visuo-motor interaction, combining the perceived changes from the environment (vision) with a rudimentary motor reaction (eye movements). The superior colliculus in the midbrain plays a central role in such rudimentary visuo-motor interactions, initiating the saccades (Hainline et al., 1984) and being connected to cortico-subcortical circuits to disengage fixation during saccades (Braddick et al., 1992). The fixation disengagement reaches functional maturity between 2 and 5 months (Hood and Atkinson, 1993), possibly reflecting the maturation of frontal cortical regions (Csibra et al., 1998). In order to track moving objects, it is necessary to (i) stabilize the target image on the retina and (ii) follow its displacements. The retinal stabilization can be achieved thanks to the optokinetic nystagmus, which subsequently needs to be inhibited in order to smoothly pursue the target's movements. In newborns, noting such ability depends on the features of the target, with smooth pursuit movements exhibited even at a few weeks of life, but only with slow and large moving targets (Phillips et al., 1997). The fact that infants can also anticipate where a moving target will go by staring at the expected location, supports that rudimentary cortical mechanisms for early visuo-motor interactions are already in place in early age (Rosander, 2007).

Eye-Hand Coordination

It is not a secret that the ability to grasp objects contributed fundamentally to render humans one of the most evolutionary successful species worldwide. Grasping is the result of complex interactions between sensory perceptions and motor control, the largest part being taken by the coordination between vision and hand movements. Such an eye-hand coordination widely permeates daily life, including object manipulation, environmental exploration, and social interaction. Without tight eye-hand links, it is doubtful that fundamental human activities like writing or driving (often given for granted, but in fact not obvious), would have evolved at such a large scale, or perhaps they would not have born at all. The existence of visuo-motor links is supported by both behavioral and neural evidence, suggesting the interaction between the ventral and the dorsal streams. Early forms of reaching and grasping emerge around the fourth month of life, supporting that the dorsal stream would be already able to coordinate the motor output in response to the visual input mediated by the ventral stream (Braddick et al., 2003). Between the sixth and ninth month, children almost compulsorily reach and grasp any object within their arm's length (Newman et al., 2001), establishing and reinforcing

rich perceptual-motor connections which will constitute scaffold for developing a broad visuo-motor neural architecture able to be activated even by less complex inputs (Pulvermüller, 1999; Martin et al., 2000). For example it has been shown that even just the observation of reaching movements activates the sensorimotor cortex in 14-month-old children with a stronger gradient as a function of older ages (Marshall et al., 2011). Similarly, visual perception of letters is associated with brain activations typical for the execution of handwriting movements (Longcamp et al., 2003), which in turn activate also visual regions typically involved in letter perception (James and Gauthier, 2006). Beyond action execution, vision can contribute also to accurate action planning, including the ability to anticipate the appropriate hand configuration to grasp a specific object (Rosenbaum et al., 1992). Typically, this ability is achieved at about 8 years (Smyth and Mason, 1997), but some delays can be encountered in presence of clinical conditions that are likely affect the interactions between the dorsal stream, ventral stream, and frontal brain areas (Braddick and Atkinson, 2013).

Not only can vision guide movements, but also motor training can affect visual perception. At the behavioral level, visuomotor training improves letter recognition in 5-year-old children (Bara and Bonneton-Botte, 2018). Similarly, handwriting improves after haptic (not visual) exploration of letters even at younger age (Bara et al., 2004), and is associated with better visual recognition of letters with respect to typing (Longcamp et al., 2005) and with better reading in general (Labat et al., 2010). At the neural level, in addition to the anatomo-functional overlap of brain regions activated by “seeing” and “doing” movements (Halje et al., 2015), already in 9-month-old children the motor components of the brain activity associated to observation of reaching actions occur earlier than the associated visual components (Southgate et al., 2009). This supports the existence of visuo-motor anticipation mechanisms based on experience-driven action understanding (Southgate et al., 2010). In addition to reaching and grasping, locomotion occupies an important position in visuo-motor coordination. In typically developing children locomotion emerges around the first year of life, strongly based on the ability of vision to provide information about the target position, possible obstacles, variations of surfaces, and edges. Thus, vision must have tight links also to the neural correlates of locomotion. Indeed already the simple observation of other children crawling or walking activates the sensorimotor cortex in 7–9 month-old children (De Klerk et al., 2015) as well as more frontal motor brain regions controlling locomotion in 14–16 month-old children (Van Elk et al., 2008), a resonance mechanism that persists in adulthood even just imagining to walk (Ionta et al., 2010).

VISUO-MOTOR NEUROPSYCHOLOGY

Conceiving bio-computational models to explain the causal link between dysfunctional neural networks and clinical phenotypes is the major challenge in neuropsychology. The following sessions offer an overview of the most common visual and visuo-motor disorders, including the possible associated neural explanations.

Broadly, the following disorders have been classified as “lower” and “higher” level deficits, even if such a sharp distinction might not reflect all the details of each disorder. The “lower-level” classification comprises disorders mostly affecting the perceptual level, with a high importance of basic mechanisms associated with eye movements and convergence. The “higher-level” classification comprises disorders affecting levels beyond visual perception and rather extending to other spheres of human competences, such as motor and cognitive skills, eventually in absence of other possibly coherent deficits.

Lower-Level Dysfunctions

Strabismus

Strabismus is one of the most common visual disorders, affecting the ability to maintain the alignment between the two eyes and therefore causing a binocularity breakdown due to the mismatch of the information provided by each eye to the visual cortex (Cullen, 2015). The importance of cortical mechanisms in the onset of strabismus is shown by the fact that, at least in monkeys, a lack of intervention at the cortical level can nullify the benefits brought by surgical treatment of the eye muscles (Pullea et al., 2018). Both in humans and other mammals, already the first weeks of life are fundamental for a proper oculomotor development leading to accurate eyes alignment (Tychsen, 2007). Due to the immaturity of V2 neurons with respect to V1 neurons, in the infant brain abnormal visual input can dramatically affect the neural wiring especially in V2 (Nakatsuka et al., 2007), the maturation of which could be misled by inappropriate experience/stimulation (Zheng et al., 2007). On this basis and in combination with the above-mentioned tight visuo-motor links, it is not surprising that the incongruent input received by V1 from the two eyes triggers a cascade of neural events ending in incongruent motor commands sent from to the oculomotor brain centers (e.g., the superior colliculus) back to the eyes (Das, 2016; Walton et al., 2017). Thus, the differential visual input of each eye would contribute to the misalignment of the eyes themselves, as supported by the inextricable relationship between sensory input and motor output (Perruchoud et al., 2014), including evidence that the onset of strabismus can derive from aberrant visual input (Chino et al., 1997). In addition to such aberrancies in the visual cortex, also disturbances in other brain areas have been linked to strabismus, such as abnormal visual-oculomotor behaviors in presence of dysfunctions in V5 and superior temporal gyrus (Mustari et al., 2008; Mustari and Ono, 2011), as well as other neuroanatomical aberrancies affecting the ventricles and corpus callosum (Ohtsuki et al., 2000). Such breakdowns in the visuo-motor loop can impair the perception of depth and also contribute to the onset of e.g., amblyopia (Sengpiel and Blakemore, 1996; Niechwiej-Szwed et al., 2019).

Oculomotor Apraxia

Optic apraxia refers to the impossibility to perform eye movements, resulting in the so-called “sticky” vision: the impossibility to voluntarily shift gaze between different objects (Pena-Casanova et al., 1985). At the neural level, bilateral lesions in a fronto-parietal network comprising the frontal eye fields are considered at the origin of oculomotor apraxia,

which therefore would not be necessarily associated strictly with dorsal stream damage (Leigh and Zee, 2015), extending to malformations/dysfunctions in the cerebellum (Shahwan et al., 2006) and midbrain (Jissendi-Tchofo et al., 2009; Merlini et al., 2010). In children, oculomotor apraxia can be present already around the 10th year of life (Tsao and Paulson, 2005), with a mean age of about 7 years and comprised between 2 and 18 years (Le Ber et al., 2003). Anatomic-functional aberrancies of the cerebellum have been repeatedly associated with oculomotor apraxia (Maria et al., 1999; Gleeson et al., 2004), with a particular responsibility for a too small cerebellar vermis (Sargent et al., 1997). The consequences of oculomotor apraxia do not remain limited to the visual domain, but rather spread on cognitive and social skills, especially in the case that oculomotor abilities are recovered too late (Kondo et al., 2007).

Amblyopia

Amblyopia can emerge when the visual input from one eye is not properly processed by the brain, which progressively develops a “preference” for the other eye. It results in atypical vision from one eye that otherwise appears organically normal (Bretas and Soriano, 2016). At the brain level, typical functional abnormalities associated with amblyopia converge in indicating V1 as the most affected brain region (Blakemore and Vital-Durand, 1986). However, the abnormal neural activity associated with amblyopia is not necessarily limited to V1, rather extending also to V2 and V3 (Barnes et al., 2001), even when V1 is normally functioning (Clavagnier et al., 2015). Interestingly, amblyopia patients present larger receptive fields in V1, V2, and V3, possibly as a consequence of the oculomotor instability of the amblyopic eye (Levin et al., 2010). Indeed there seem to be a sort of propagation of dysfunctional neural dynamics from V1 up to V5 (Barnes et al., 2001), which would result in specific deficits in extrastriate functions, including global motion (Simmers et al., 2003) or contrast-based contours (Wong et al., 2001).

Akinetopsia

Our ability to perceive motion allows us to distinguish objects from the background and to move in a three-dimensional world (Barton, 2011). Commonly called also motion blindness, akinetopsia refers to the impossibility to detect moving objects, in absence of scotoma (Zihl et al., 1983), while other low-level aspects like color or shape are normally detected (Zeki, 1991). Typically associated with an extrastriate brain lesion (Zihl et al., 1983; Cooper et al., 2012; Otsuka-Hirota et al., 2014), akinetopsia can indeed be experimentally induced by inhibiting V5 (Beckers and Hömberg, 1992), as well as V1 but at a smaller degree and with specific timing with respect to the visual stimulus (Beckers and Hömberg, 1992). This is in line with the observation that sensitivity to motion can survive cortical blindness (Ruffieux et al., 2016), also in children that present a congenital, but not acquired, lesion of V1 (Tinelli et al., 2013). While blindness to first-order motion (e.g., luminance-based) would result from lesions in V2/V3, blindness to second-order motion (e.g., contrast-based) would derive from lesions in V4/V5 (Cowey et al., 2006). A particular case of motion blindness is represented by the “form-from-motion” blindness, referring to

the impossibility to detect forms on the basis of visual motion (Cowey and Vaina, 2000). Indeed form-from-motion blindness with and without akinetopsia are neurally dissociable, being the former associated with lesions in V5 and lateral occipital cortex and the latter with occipito-temporal regions (Blanke et al., 2007). Even if chronic cases have been reported (Cooper et al., 2012), akinetopsia seems a rather transient condition (Shipp et al., 1994), suggesting the existence of functionally neuroplastic changes able to establish alternative neural interactions to restore sensitivity to visual motion. Such a relative ease to naturally react to akinetopsia makes it difficult to detect, especially in populations characterized by high neural plasticity like children, where in fact akinetopsia is relatively rare and usually present in combination with the Alice in Wonderland syndrome as a results of encephalitis (Naarden et al., 2019).

Higher-Level Dysfunctions

Optic Ataxia

Originally described by Rudolph Bálint in 1909 as part of a more complex syndrome (Rudolph Bálint, 1909), optic ataxia refers to the incapacity to perform accurate visually-guided movements, in absence of general motor impairments (Moreaud, 2003). Letting patients misplace the fork outside the plate, grasp a coffee mug from its body instead of its handle, or point to the wrong button on a computer keyboard, optic ataxia is considered the typical visuo-motor integration disorder (Teixeira et al., 2014). Not strictly limited to visuo-motor behaviors of the upper limb (Evans et al., 2013), it can emerge as early as in children aged between 5 (Dutton, 2003) and 10 years (Drummond and Dutton, 2007). Possibly as a consequence of premature birth (Dutton, 2013), optic ataxia has a confirmed association with aberrancies in the (occipital-parietal) dorsal stream (Philip et al., 2016). Indeed, the most accepted neural underpinnings of optic ataxia are comprised within the dorsal stream (Schindler et al., 2004), possibly also in interaction with the ventral stream (Himmelbach and Karnath, 2005). Further investigations reported that optic ataxia affects mainly the peripheral vision (Pisella et al., 2009) and is especially evident in contralesional visuo-motor tasks (Gaveau et al., 2008). This suggests that optic ataxia should not be considered as a unitary deficit, but rather presents various degrees and specifications as a function of the lesioned dorsal stream module responsible to coordinate visual perception and action. Nevertheless, recent evidence is starting to challenge such a sharp dissociation between perception and action in optic ataxia (Rossetti and Pisella, 2018). In particular, optic ataxia would derive from dorsal stream deficits in integrating multimodal sensory input (Jackson, 2010), it can be stimulus/task-specific (Hesse et al., 2014), and it can be bound to specific visuo-motor neurons located in different regions of the dorsal stream and beyond (Cooper and O’sullivan, 2016), especially the premotor cortex (Battaglia-Mayer and Caminiti, 2002) and a parietal-precuneus pathway (Teixeira et al., 2014).

Cerebral Visual Impairment

As one of the most common causes of visual impairment of cortical origin, cerebral visual impairment (CVI) can result

from early brain damage, including a potentially large panel of correlated deficits beyond vision due to damages of the dorsal stream, the ventral stream, or both (Bennett et al., 2020). Behaviorally, it is possible to detect CVI by means of dedicated questionnaires (Gorrie et al., 2019; Fazzi and Micheletti, 2020). At the neural level, a relatively early detection of CVI is based on the analysis of visual evoked potentials which, already at 6 months of age, can appear abnormal and therefore suggest the presence of CVI (Mercuri et al., 1997b), further depending on the size (Mercuri et al., 1998) and location (Mercuri et al., 1997a) of the brain lesion. In particular, the basal ganglia seem to play a central role in coordinating the information exchanges between the eyes and the visual cortex, as well as in facilitating neural plasticity at the cortical level (Mikellidou et al., 2019), possibly resulting in aberrant patterns of anatomo-functional connectivity between different brain regions (Muñoz-Moreno et al., 2016; Bathelt et al., 2020). It is anyways important to note that CVI can impair a full range of competences at different levels, including purely visual skills (visual field, motion sensitivity, visual exploration) as well as attention, memory, and visuo-motor coordination (Lueck et al., 2019). This is the main reason why current trends in neuro-ophthalmology highlight the importance of considering each patient as an individual case that should be evaluated on the basis of a personalized and multidisciplinary assessment combining ophthalmology, neuropsychology, and pedagogy (Ortibus et al., 2019).

Dorsal Stream Vulnerability

As already outlined, the dorsal stream is considered the main neural architecture processing spatial aspects of vision and their translation into relevant information for functions beyond the mere sight. Converging evidence supports that the dorsal stream is more vulnerable than the ventral stream to developmental disorders (Grinter et al., 2010), due to genetic or contextual factors (Atkinson, 2017) as well as interventional approaches (Tonks et al., 2019). Possibly leading to cognitive decline (Ricci et al., 2015), attentional/visuo-spatial deficits (Tonks et al., 2019), and visuo-motor impairments (Atkinson and Braddick, 2011), the dorsal stream vulnerability can start in early age and keeps affecting the individual competences from early childhood across the life span (Sciberras-Lim and Lambert, 2017). Nevertheless, recent findings are starting to challenge this view, by arguing that dorsal stream vulnerability might be stimulus-specific rather than a general dysfunction (Joshi et al., 2020), as shown for example by the relatively preserved motion sensitivity in amblyopia (Hamm et al., 2014). Beyond the stimulus-specificity, such a controversy might result also from task-specificity since, for example, some dorsal stream functions (e.g., time estimation and attentional tasks) seem more sensitive to developmental disorders than others (e.g., numerical discrimination or mapping). In consideration of such a variability among stimuli and tasks, it is clear that to evaluate a wide range of symptoms like those related to dorsal stream vulnerability implies the need of using multidimensional scales for evaluating dorsal stream vulnerability (Atkinson et al., 2002).

Developmental Coordination Disorder

The diagnosis of Developmental Coordination Disorder (DCD) is based on the presence of motor impairments in absence of other neuropsychological deficits able to explain patient's poor motor performance (cerebral palsy, neurodegeneration, traumatic brain injuries, etc.) (Blank et al., 2019). The characteristics of DCD include impaired control of ocular, postural, and manual tasks, as well as motor imagery (Adams et al., 2014). One of the possible interpretations of DCD explains the disorder as the result of breakdowns in a visuo-motor matching system which would allow to perform movements on the basis of observing the same movements performed by somebody else (Werner et al., 2012). Such a breakdown would affect in particular the ability to process and exploit the temporal binding between vision and movements (Nobusako et al., 2018). The visuo-motor interpretation of DCD is in line with evidence that DCD patients exhibit impaired visuo-motor skills (Reynolds et al., 2017) and decreased brain activation in regions typically involved in visuo-motor imitation (Licari et al., 2015) and action planning (Reynolds et al., 2019). In particular, even if a large consensus has not been reached yet (Brown-Lum and Zwicker, 2015), it seems that the brain dysfunctions associated with DCD are mainly located in associative regions of the parietal and frontal lobe particularly important for visually-based action imitation (Biotteau et al., 2016). In sum, despite the little number of studies and the large variability of their results, there is a tendency to consider DCD as a visuo-motor integration deficit specifically affecting the neural network responsible for visually interpreting actions performed by other people and exploit such information for guiding self-produced movements. However, further studies are required and the present conclusions have to be regarded with caution.

Prosopagnosia

The ability to recognize faces is one of the most important abilities in the human world. The centrality of this function is reflected in the fact that the brain dedicates a specific neural substrate to process face-like visual input (Zeugin et al., 2020), with a particular emphasis on the fusiform face area in the (ventral stream) inferior temporal cortex (Kanwisher et al., 1997). Prosopagnosia refers to the inability to recognize faces (Mayer and Rossion, 2007) associated with occipito-temporal brain activity (Dalrymple et al., 2014) and, in particular, with bilateral lesion of the fusiform face area (Grüter et al., 2008). It is dissociated from other object-recognition deficits as, for example, there are cases in which in consequence of a bilateral fusiform lesion patients become unable to recognize faces while their performance in object recognition remains at good levels (Moscovitch et al., 1997). Since prosopagnosic people are largely unaware of their deficit, prosopagnosia can dramatically affect the cognitive development of otherwise typically growing children (Schmalzl et al., 2008), including the preference for social identification on the basis of whole-body configuration instead of facial features (Wilson et al., 2010). This conditions can trigger a cascade of aversive events and behaviors also in daily contexts like schools, where both teachers and colleagues would

not detect the prosopagnosic deficit and therefore might put disproportionate reactions in place (Wilson et al., 2010). To prevent and possibly overcome this risk, at present there are strong trends toward the development of specific test to assess face perception abilities in children, such as the Dartmouth Database of Children's Faces (Dalrymple et al., 2013) and the Cambridge Face Memory Test for Children (Croydon et al., 2014).

Somatoparaphrenia

The ability to recognize our own body also plays a central role in adaptive behaviors and consciousness (Ionta et al., 2013). Somatoparaphrenia refers to the inability to identify one's own body part as belonging to one self, both at the subjective conscious (Invernizzi et al., 2013) and objective physiological levels (Romano et al., 2014). Despite its psychiatric component (Feinberg and Venneri, 2014), somatoparaphrenia is largely associated with unilateral lesions, mostly in the right hemisphere and therefore affecting the left side of the body (Vallar and Ronchi, 2009). At a more specific neural level the available evidence is controversial, with clinical observations reporting damages in either the dorsal or the ventral stream, as well as other brain regions. Thus, different studies proposed that somatoparaphrenia would derive from lesions in the posterior insula (Baier and Karnath, 2008), supramarginal gyrus (Feinberg et al., 1990), orbito-frontal regions (Feinberg et al., 2010), posterior superior temporal cortex (Vallar and Ronchi, 2009). In addition, recent investigations highlighted the importance of more complex fronto-temporal-parietal cortical networks as well as subcortical circuits (Gandola et al., 2012). Even if somatoparaphrenia is commonly associated with hemispatial neglect, it can be present also in isolation and associated with specific subcortical lesions, comprising the basal ganglia, thalamus, and internal capsule (Invernizzi et al., 2013). Interestingly when somatoparaphrenic patients observe the misrecognized body part in a mirror (as from a third-person perspective), their self-misattribution decreases (Fotopoulou et al., 2011). Already rarely detected in adults, possibly due to its comorbidity with hemispatial neglect and its confusion with asomatognosia, evidence of somatoparaphrenia in children is even more scarce. However, a study implementing a neuroinvestigation technique with high spatial resolution (electrocorticography) in awake humans, reported that following abnormal neural firing in the right occipito-temporo-parietal cortex, a 10-year-old child reported somatoparaphrenic symptoms, being unable to recognize his left hand (Heydrich et al., 2011). Altogether, it seems that the small numbers to somatoparaphrenic reporting reflects a general lack of episodes spontaneously mentioned by the patients together with the confusion with other pathologies by the evaluators. For this reasons, it would be important to explicitly assess somatoparaphrenic symptoms using structured interviews (Brandt et al., 2005) and/or standardized scales especially in developing age.

Hemispatial Neglect

The absence of perception and action in half of the sensory fields and peri-personal space, respectively, defines the hemispatial neglect. Patients suffering from this syndrome do not perceive sensory stimuli in any modality from the neglected hemifield and do not perform movements in that hemi-peri-personal space. The traditional test to assess neglect is the line bisection task, in which patients are presented with a paper sheet with a number of short lines distributed all over a paper sheet. Typically, when patients are asked to draw a line over each short line (bisection), they mark only half of the lines (those located in the non-neglected hemifield). At the neural level, hemispatial neglect seems to derive from dysfunctions in the right inferior parietal lobule, possibly in association with deteriorated input from the ventral stream (Milner and Goodale, 2008) or with impaired ventro-fronto-parietal circuit distinct from the traditional dorsal stream (Husain and Nachev, 2007). However, hemispatial neglect can emerge also following lesions of the frontal cortex (Husain and Kennard, 1996), basal ganglia or thalamus (Mort et al., 2003), as well as from lesions of white matter pathways connecting the parietal and frontal cortex (Bartolomeo et al., 2007). Altogether, it seems that hemispatial neglect may be the result of a lesions in a large-scale cortico-subcortical network, possibly implicated in attention-related abilities. Interestingly, while most intervention protocols eventually produce only temporary improvements, the most efficient and relatively long-lasting treatment is based on the use of prism adaptation (Rossetti et al., 1998). In particular, the visual distortion brought by wearing prism lenses would trigger the activation of otherwise silent visuo-motor circuits as valid alternative neural pathways to allow visuo-motor coordination, with benefit spreading also in the cognitive domain (Rossetti et al., 1998). Even if hemispatial neglect is commonly associated with adult and elderly patients, also children can be affected, and not necessarily only in the visual domain (Martin and Trauner, 2019). Cases of hemispatial neglect have been reported for children as young as 3-year-old (Thompson et al., 1991), 6-year-old (Ferro et al., 1984), and above (Hausmann et al., 2003; Marsh et al., 2009). Actually, also at 6 months after birth, children with pre- or post-natal brain damage can exhibit otherwise unmotivated preference for interacting with objects located in the hemi-peri-personal space ipsilateral to a unilateral lesion in the left or right hemisphere (Trauner, 2003). Most of the studies indicate that children can relatively quickly recover from neglect symptoms within a few weeks (Kleinman et al., 2010) or months (Thompson et al., 1991) after a stroke. Even children that suffered from a perinatal stroke, especially in the right hemisphere, can present hemi-neglect-like symptoms in the left hemi-field and peri-personal space, including visuo-motor deficits (Vicari et al., 1998), reaching and grasping (Trauner, 2003), as well as visual cancellation and manual exploration (Thareja et al., 2012). These studies further showed that, in contrast with the typical right-hemispheric dominance of hemi-spatial neglect in adults, in children a more dramatic bilateral neglect can result from a left-hemispheric lesion (Trauner, 2003; Thareja et al., 2012), whose resolution might require maturation up to adolescence or adulthood (Yousefian et al., 2015).

FINAL REMARKS

Understanding the behavioral and neural fundaments of the complex interaction between vision and other sphere of human life is the prerequisite for better targeted interventional procedures in case of deficits, as well as for more efficient training programs in typically developing populations. As a very general overview, the present paper summarizes some of the most relevant evidence about the neural basis of vision and associated abilities in development and beyond. With the aim of constituting a first-glance reference for researchers and clinicians interested in vision and visuo-motor integration, this review hopes to guide and trigger further investigations toward more specific publications in case of specific interests.

Establishing the neural correlates of aberrant behaviors helps identifying the neural networks responsible for a given function which, in turn, can boost the development of more effective training and rehabilitation protocols. Accordingly, the knowledge summarized here sustains the importance of adopting a systemic approach even in the evaluation of the impact of

supposedly purely visual deficits, which indeed can affect also motor skills, cognition, social skills, and emotional processing. Addressing such a complexity is the fundamental requirement of current implementations of systemic approaches for visually-related training in typical conditions or in response to visual disorders, including virtual reality (Adams et al., 2018; Choi et al., 2021), robotics (Mirkowski et al., 2019; Zhexenova et al., 2020), and touch screen technology (Aslam et al., 2016; Sheehan and Uttal, 2016; Dalecki et al., 2019).

AUTHOR CONTRIBUTIONS

SI conceived the work, performed the literature analysis, and wrote the manuscript.

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Early Blindness Limits the Head-Trunk Coordination Development for Horizontal Reorientation

Davide Esposito^{1,2}, Alice Bollini¹ and Monica Gori^{1*}

¹ Unit for Visually Impaired People, Istituto Italiano di Tecnologia, Genoa, Italy, ² DIBRIS, Università di Genova, Genoa, Italy

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Silvio Ionta,
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*Correspondence:

Monica Gori
monica.gori@iit.it

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During locomotion, goal-directed orientation movements in the horizontal plane require a high degree of head-trunk coordination. This coordination is acquired during childhood. Since early visual loss is linked to motor control deficits, we hypothesize that it may also affect the development of head-trunk coordination for horizontal rotations. However, no direct evidence exists about such a deficit. To assess this hypothesis, we tested early blind and sighted individuals on dynamic sound alignment through a head-pointing task with sounds delivered in acoustic virtual reality. Participants could perform the head-pointing with no constraints, or they were asked to immobilize their trunk voluntarily. Kinematics of head and trunk were assessed individually and with respect to each other, together with spatial task performance. Results indicated a head-trunk coordination deficit in the early blind group; yet, they could dampen their trunk movements so as not to let their coordination deficit affect spatial performance. This result highlights the role of vision in the development of head-trunk coordination for goal-directed horizontal rotations. It also calls for clarification on the impact of the blindness-related head-trunk coordination deficit on the performance of more complex tasks akin to daily life activities such as steering during locomotion or reaching to targets placed sideways.

Keywords: audiomotor integration, egocentric reference frame, head-trunk coordination, early blindness, virtual reality, subjective straight-ahead

INTRODUCTION

How we move is closely linked to how we perceive. This connection is supported by several experimental pieces of evidence showing abnormal motor patterns in people with different types of sensory disabilities, like visual (Haibach et al., 2014), hearing (Vitkovic et al., 2016), and vestibular (Inoue et al., 2013) impairments. However, such evidence refers to passive postural balance and gross-motor abilities that involve the control of limbs. Much less is known about the influence of sensory impairments on the coordination of head and trunk goal-oriented movements. Yet, head-trunk coordination is of primary importance for accomplishing fundamental tasks, such as moving (re-orienting) to new target locations (Hollands et al., 2001). In fact, it has been shown that head-only rotations are used by the brain to prompt the body to steer toward the new head direction during locomotion (Patla et al., 1999). Interestingly, it is believed this motor pattern provides the brain with a stable spatial reference frame for body reorientation (Grasso et al., 1996).

Despite its importance, the coordination of head and trunk movements does not seem innate but is instead acquired through development. Assaiante et al. (2005) showed that children up to five-years-old move head and trunk together to some degree in postural, locomotion, and reaching tasks (Assaiante, 1998). In the same study, the researchers named this motor behavior “*en-bloc*,” in contrast to the adults’ “articulated” strategy introduced above, where head and trunk unbind and move independently. Moving head and trunk together as children do is supposed to simplify motor control by reducing the number of degrees of motion involved in a movement (Assaiante et al., 2005). Nevertheless, a significant amount of information for one’s horizontal orientation estimation is provided by proprioceptive inputs from the neck (Warren, 1998; Pettorossi and Schieppati, 2014); so, turning head and trunk together instead of coordinating the two body parts would impoverish that information source. If the available information for horizontal orientation estimation is poorer, one can hypothesize that the estimation quality will be poorer too.

Given that sensory disabilities affect several aspects of motor development, and given the developmental nature of head-trunk coordination, sensory impairments may affect head-trunk coordination as well. For visual loss, this is certainly the case. As infants, totally blind individuals show a clear delay in head control development (Precht et al., 2001). As adults, blind people lock head and trunk movements in postural balance (Easton et al., 1998; Schmid et al., 2007; Alotaibi et al., 2016). Such results recall the abovementioned “*en-bloc*” strategy, typical of sighted children whose head-trunk coordination is incomplete. Sighted children use this strategy in both postural balance and horizontal orienting movements (Assaiante et al., 2005); therefore, such coupling may hold in early blind adults too. If such a strategy were used by the adult blind population for horizontal reorientation, it should reduce proprioceptive inputs from the neck. One may therefore expect blind people to perform poorly in tasks that require horizontal head turns, such as horizontal sound localization by head pointing. Instead, scientific evidence shows that they perform as well as, or even better than, sighted individuals in this task (Lessard et al., 1998; Röder et al., 1999; Collignon et al., 2009; Lewald, 2013).

The apparent contradiction between spatial and kinematic performance among blind people may be explained in two ways. On the one hand, visual loss does not cause head-trunk coordination issues for goal-directed horizontal rotation. On the other hand, head-trunk coordination may be impaired in blindness, but simple head-pointing tasks with none or passive-only constraints on trunk movements may not challenge head-trunk coordination enough to affect spatial performance. However, to the best of our knowledge, no prior study has directly tested blind people’s head-trunk coordination in goal-directed horizontal rotations, nor their horizontal audio-spatial performance while their head-trunk coordination is challenged. Given that early blind adults show child-like head-trunk coordination for postural tasks (Easton et al., 1998; Schmid et al., 2007; Alotaibi et al., 2016), and given that in children the head-trunk coordination strategy is similar in postural tasks and in tasks requiring goal-directed horizontal rotations

(Assaiante et al., 2005), we hypothesize the early blind adults would also show child-like head-trunk coordination in a goal-directed horizontal rotation task.

We developed a dynamic sound alignment task on an acoustic virtual reality (AVR) platform made expressly to test this hypothesis. AVR environments are handy tools for defining complex tasks involving auditory localization in the horizontal plane because they give results similar to those with real speakers (Wenzel et al., 1993). Furthermore, they provide portable setups that guarantee more control over the sound position relative to the ears and inherently provide kinematic data about the tracked body parts. In order to test our hypothesis, AVR allowed us to define an experimental task based on sound localization in the horizontal plane via dynamic head-pointing, with or without acoustic feedback for trunk movements. With their kinematic profiles, we could directly evaluate participants’ head-trunk coordination. At the same time, with the sound localization task we could check the extent to which different degrees of head-trunk coordination relate to audio-spatial performance. In order to challenge participants’ head-trunk coordination, we set a head-trunk coordination constraint to demand voluntary trunk immobilization by means of acoustic feedback for trunk movements and explicit instructions. This condition was paired with another, where the head-trunk coordination was spontaneous, without feedback. Doing so, we could expose behavioral differences in horizontal sound alignment by head-pointing with and without demanding head-trunk coordination.

We tested typical sighted and early blind participants on our AVR platform. Following our hypothesis we predicted that if early visual deprivation affected the head-trunk coordination for horizontal rotations, early blind people would differ from sighted controls when demanded to coordinate head and trunk in kinematic behavior and, if the impairment is large enough, also in spatial performance.

METHODS AND ANALYSIS

Participants

In total, 21 individuals, 10 congenital blind (3 males, 7 females, age = 33.2 ± 3.19 years old, the clinical details of the participants’ pathologies are reported in **Table 1**) and 11 sighted individuals (6 males, 5 females, age = 31.27 ± 3.92 years old) were involved in the study. All of them were enrolled by local contacts in Genoa. Informed consent was obtained from all participants. The study followed the Helsinki Declaration’s tenets and was approved by the ethics committee of the local health service (Comitato Etico, ASL 3, Genova).

Physical Experimental Setting

The AVR environment created for this experiment was developed with the game engine Unity 3D. The spatial blending of sounds was made using the *resonance* package (Google, 2018). The sound was delivered via commercially available BOSE® over-ear headphones. For the purpose of the kinematic assessment, participants’ head and trunk movements were tracked. The participants’ head rotations were tracked by the head-mounted

TABLE 1 | Clinical details of blind participants.

	Gender	Age	Pathology	Blindness onset	Residual vision
P1	F	32	Retinopathy	Before birth	No vision
P2	F	20	Retinopathy	Before birth	Lights and shadows
P3	F	29	Retinopathy	Before birth	No vision
P4	M	27	Leber's amaurosi	Since birth	No vision
P5	F	26	Glaucoma and retinal detachment	Before birth	No vision
P6	M	46	Leber's disease	Before birth	No vision
P7	M	52	Unknown	Before birth	Lights and shadows
P8	F	30	Retinitis pigmentosa	Since birth	Lights and shadows
P9	F	28	Microphthalmia	Since birth	No vision
P10	F	42	Retinopathy	Since birth	No vision

display (HMD) itself, at a sampling rate of 90 Hz, which is the frequency of Unity's main loop. The trunk rotations were tracked by an LG® google nexus 4 smartphone used as a wireless inertial measurement unit thanks to the app HyperIMU (Ianovir, 2019), with a nominal sampling rate of 100 Hz. Both sensors have a resolution of 0.1° . The incoming samples were asynchronously collected via an User Datagram Protocol (UDP) socket, stacked and averaged at the frequency of 90 Hz. The smartphone was fixed to participants' backs using a custom-made harness. In the context of AVR, the HMD screens were blank, so the virtual reality headset (HTC® VIVE) was uniquely used to track the participants' head movements. During the experiment, participants were seated and their trunk was free to rotate (**Figure 1**). Unity's refresh rate of the physics engine was kept at the standard value of 30 Hz to maintain a good tradeoff between performance and computational cost. The HTC Vive headset was chosen because its tracking system is reliable, accurate, validated for scientific research (Le Chénéchal and Chatel-Goldman, 2018; Niehorster et al., 2017; Verdelet et al., 2019) and comes with default Unity 3D integration. The smartphone IMU was preferred over HTC Vive trackers to reduce the risk of tracking loss for whatever reason, which is a known performance issue for the Vive system (Niehorster et al., 2017; Verdelet et al., 2019). IMUs, instead, are subject to drift. Recalibration was performed before starting a new condition and after approximately 10 trials to compensate for the drift effect.

Virtual Experimental Setting

The AVR platform we developed defines four goal-directed steering tasks in a first-person perspective, described as archery-like games, based on the same virtual environment. The virtual environment's absolute reference frame is aligned to the participant's seat via the standard calibration phase for the HTC VIVE. The unit of measure of length is the unity unit (uu). Objects' sizes and distances have been designed to match the uu with the meter; therefore, the meter will be used to describe the spatial parameters. The camera view is 1.7 m above the ground and its position in the virtual environment mimics the arrow position. The arrow can be in two states only: loaded and shot. In the loaded state, the arrow appears at the origin of the virtual

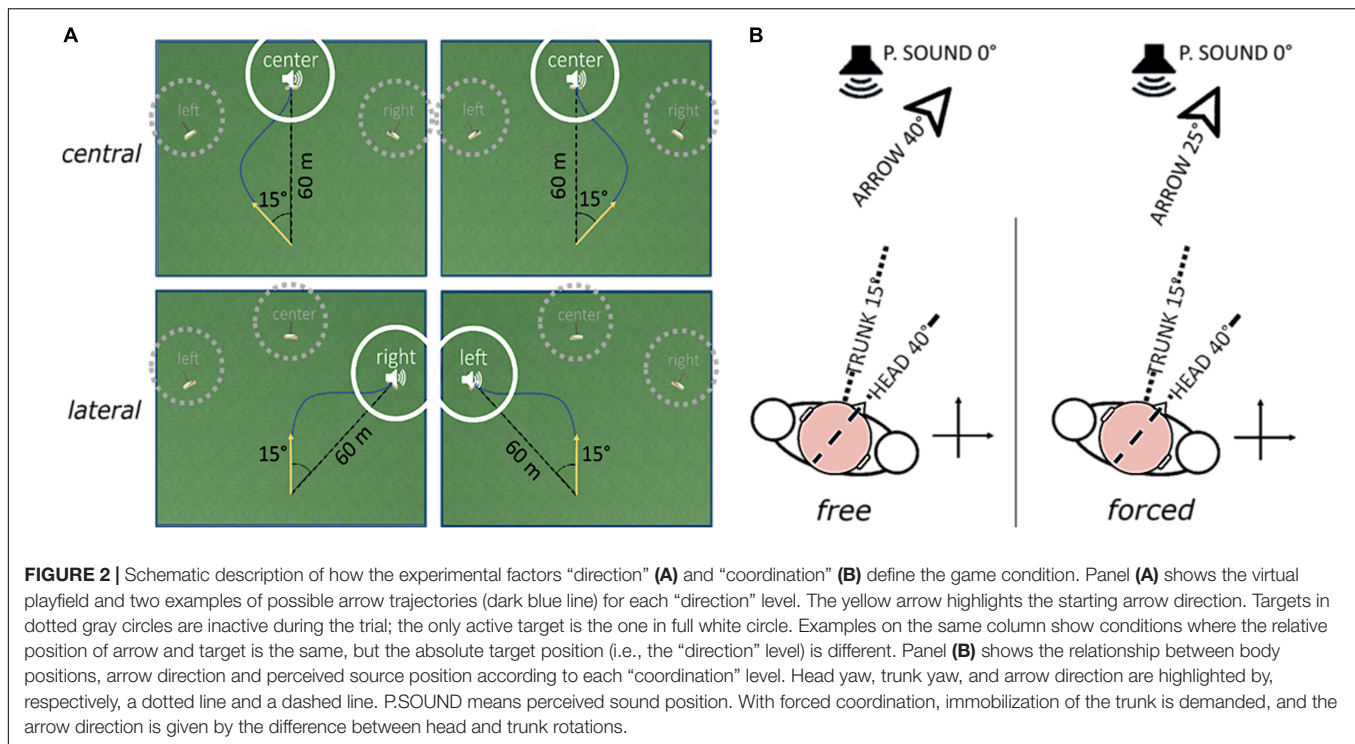


FIGURE 1 | Hardware and room setting during an experimental session. The participant wears the VR headset, the headphones and a custom-made harness to keep the smartphone on his/her back. During each trial, the participant sits and his/her back is free to rotate.

environment's absolute reference frame; it does not move, but it can rotate around the vertical axis. The transition from loaded to shot state is automatic. It happens when the arrow orientation lies inside a trigger window for a time span randomly chosen from 1 to 3 s. The arrow, once shot, advances at a fixed velocity of $10 \frac{m}{s}$ in the horizontal plane. The target is a source of intermittent pink noise with 5 Hz duty cycle; its spatial attenuation follows an isotropic logarithmic function. It is 60 m distant from the starting point and can appear at three absolute angles: -15° , 0° , $+15^\circ$. Regardless of the task, trigger window, and target centers are always shifted $\pm 15^\circ$ apart from each other. **Figure 2A** displays the virtual environment, together with the set of possible trigger window-target geometrical configurations.

Tasks Description

The four above-mentioned archery-like, first-person perspective, goal-directed steering tasks are actually four conditions of one base task, derived by four different parameterizations. The base task consists of leading the arrow, whose initial trajectory is by-design 15° apart from the target, toward the target itself. The arrow flight direction depends on a combination of head and trunk rotations around the vertical axis, and the participants' goal is to hit the target center.



The conditions are defined according to two factors: “direction” and “coordination” (Figure 2).

The factor “coordination” rules the combination of head and trunk movements leading the arrow direction in the virtual space. Its two levels are *free* and *forced*. In the *free* level, the experiment baseline, the arrow rotation mimics the head yaw uniquely and participants are told by the experimenter to move freely, only caring about hitting the target. In the *forced* condition, the test, the trunk is used as virtual environment’s reference frame. In this way, the arrow rotation mimics the difference between head and trunk yaws, and the relative target-to-trunk rotation is kept constant throughout the trial. This condition was designed to discourage trunk rotations implicitly. Moreover, participants were explicitly asked by the experimenter to voluntarily immobilize their trunk as much as they can. The comparison between these two levels, one without coordination demand and the other with active coordination demand, is the method used to assess the relationship between early visual impairment and head-trunk coordination.

The factor “direction” sets the relative initial position of the target concerning the arrow. Its two levels are *central* and *lateral*. In the *central* level, the target is aligned with the participant’s straight-ahead and the starting position of the arrow is 15° to the side, randomly alternated between rightward and leftward. In the *lateral* level, the target is positioned at 15° from the participant’s straight-ahead, randomly alternated between rightward and leftward, and the starting position of the arrow is 0° (the participant starts with head and trunk aligned to the straight-ahead direction). The distinction between *central* and *lateral* targets was implemented because previous research on auditory localization in the horizontal plane by

head-pointing showed better spatial performance in frontal than in eccentric stimuli (Makous and Middlebrooks, 1990; Lewald et al., 2000; Occhigrossi et al., 2021). By doing so, we aimed at evaluating possible differences in motor behavior that could explain those findings.

Each trial is made of two steps: positioning and execution. In the first step, positioning, the arrow is yet to be shot and the participant places the arrow in the trigger as mentioned above the window. Acoustic feedback is provided to help participants find the required starting orientation. This is an intermittent pure tone whose pitch is tuned by the angular distance between arrow and trigger window; the shorter the distance, the higher the pitch. The arrow shooting is announced by the interruption of the intermittent pure tone feedback and the reproduction of an arrow-shot-like sound. Data collected in this first step are not in the interest of this study. The second step, execution, is where the arrow moves and the task is accomplished. The trial end is notified by a prerecorded soundclip of an arrow hitting a wall if the target is hit, or the sound of a windblow if the target is missed.

Experimental Procedure

At the participants’ arrival, they were given the following instructions: “Imagine you are on an arrow. Once shot, it will fly at a constant speed and you will control its direction only by moving a part of your body as I will tell you. Your goal is to drive the arrow toward the sound you will hear, which will correspond to the target. There are three possible target positions.” Then, they were blindfolded and introduced to the virtual platform by tactile exploration of a scaled plastic model of the environment; the experimenter made them track with a finger four plausible arrow paths, two for each “direction” condition (e.g., blue curved

lines in **Figure 2A**). To let the participants familiarize themselves with the platform, a very short training session was performed, made of no more than eight trials in the baseline condition, *free* coordination. To make sure participants could exploit the audio feedback, the experimenter guided the participants' head movements by hand during the first three/four familiarization trials. In the remaining trials, participants tried to drive the arrow on their own. For the training, only one "direction" level was randomly chosen, counterbalanced among participants.

Each experiment consisted of four runs, in randomized order and counterbalanced among participants. Each run was made of twenty trials. Before proceeding with a run, the corresponding requirements were explained to the participant by the experimenter. Breaks were allowed at any time according to participants' needs. The whole experiment lasted approximately 30 min with no breaks.

Data Analysis

Two behavioral aspects were evaluated in this experiment: task-related performance and motor behavior. The task-related performance was evaluated by means of accuracy and precision in hitting the target center. The arrow hit-point distance from target center (i.e., final error) was attributed a sign according to a target-based coordinate system. Specifically, given that the line joining target and absolute coordinates' origin splits the virtual environment in two hemispaces, if the arrow end-point lied in the same hemisphere as its initial trajectory, it would be positive. Otherwise, it would be negative. Then, data distributions from each condition were tested for normality using the Lilliefors test (Lilliefors, 1967). For both the distributions' high non-normality rate and the small groups' sample sizes, we decided to use non-parametric statistics. Consequently, the accuracy was computed as the median of the by-trial final error, and the precision as the inter-quartile range (IQR) of the by-trial final error.

The motor behavior evaluation was based on the analysis of head and trunk yaws (deg) collected during trials' execution step. Raw signals were acquired at Unity's main loop refresh rate, which is 90 Hz only approximately. To compensate for sampling jitter and missing data points, the signals were resampled at 90 Hz, then smoothed using an 18-samples moving average window. Further analyses were performed on yaw jerks (variation of angular acceleration, $\text{deg}\cdot\text{s}^{-3}$) in each execution step. Two measures were used to evaluate the motor behavior: root mean square (RMS) of the trunk yaw jerk signal, in brief trunk RMS, used to quantify how much it was moved; and the amplitude of cross-correlation peak between head and trunk yaw jerks, in brief cross-peak, used to quantify the similarity between head and trunk movements. Again, the median was used to aggregate by-trial measures.

Since data points in kinematic signals were not independent and identically distributed, head and trunk yaw jerks were prewhitened before computing cross-correlation (Dean and Dunsmuir, 2016). The **Supplementary Material** contains a full methodology description.

Accuracy, precision, RMS and cross-peak were analyzed using a three-ways $2 \times 2 \times 2$ repeated-measures ANOVA on aligned rank transformed data, and ART ANOVA

(Wobbrock et al., 2011) with "group" as a between-subjects factor (blind and sighted), and "direction" (*central* and *lateral*) and "coordination" (*free* and *forced*) as within-subjects factors. *Post-hoc* comparisons were performed via Wilcoxon test for within-group comparisons and Mann-Whitney test for between-group comparisons. In case the ART ANOVA returned significant interaction effects, *post-hoc* comparisons were performed between the interaction levels with one main level in common, and *P* values were Bonferroni corrected. Standardized effect sizes were computed along with the unstandardized tests. Partial eta squared is provided as standardized effect size for the ART ANOVA. Rank biserial correlation and its confidence interval are provided as standardized effect size for non-parametric *post-hoc* tests.

Kinematic data resampling, smoothing and differentiation, prewhitening, estimation of cross-peaks and RMS were made with the software MATLAB r2020a. ART ANOVA and *post-hoc* analyses were made with the software R. ART ANOVA were made with the package ARTool (Wobbrock et al., 2011). The final dataset can be found in the Zenodo repository at the link <http://doi.org/10.5281/zenodo.4707477>. Raw or intermediate datasets generated during the current study and code used for the analysis are available from the corresponding author on reasonable request.

RESULTS

Data collected with *forced* coordination were compared to those collected with *free* coordination to evaluate the ability of early blind individuals and sighted controls to localize dynamic sounds by head-pointing while self-immobilizing their trunk on both kinematics and spatial performance standpoints. The distinction between *central* and *lateral* direction levels was maintained in the analysis to assess behavioral differences between frontal and eccentric targets, as previously found for auditory localization by head-pointing (Lewald et al., 2000; Occhigrossi et al., 2021). Results are reviewed separately for each measure. Hypotheses for each measure are expressed in the corresponding subsection. All the results of the ANOVA tests are reported in **Table 2**. Data boxplots are reported in **Figure 3**.

Kinematics

Kinematic behavior was assessed in terms of trunk jerk RMS and cross-peak. The trunk jerk RMS is a measure of the amount of movement. A larger RMS means participants moved the trunk more. The cross-peak is a measure of similarity between the head and trunk angular jerk. A larger peak means more similar head and trunk movements.

Trunk Jerk RMS

If people tried to immobilize their trunk, their trunk RMS would be larger in *free* than in *forced* conditions; however, if early blind people struggled at it, their trunk RMS in *forced* conditions would be larger than that of sighted participants. If having targets straight-ahead facilitated trunk immobilization, the trunk RMS would be larger in the *lateral* than in the

TABLE 2 | Results of the ANOVA tests for each of the four computed measures.

Effect	TRUNK JERK RMS			XCORR PEAKS			ACCURACY			PRECISION		
	F(1,19)	Pr(> F)	p.eta ²	F(1,19)	Pr(> F)	p.eta ²	F(1,20)	Pr(> F)	p.eta ²	F(1,20)	Pr(> F)	p.eta ²
group	18.40	0.000***	0.49	5.23	0.034*	0.22	0.39	0.539	0.02	0.19	0.665	0.01
direction	0.05	0.826	0.00	1.08	0.312	0.05	3.28	0.085	0.14	1.08	0.312	0.05
coordination	6.02	0.024*	0.24	12.02	0.003**	0.39	0.90	0.355	0.04	0.02	0.881	0.00
group:direction	0.32	0.578	0.02	1.71	0.206	0.08	0.41	0.527	0.02	0.00	0.980	0.00
group:coordination	3.59	0.074	0.16	9.04	0.007**	0.32	1.64	0.214	0.08	6.87	0.016*	0.26
direction:coordination	0.97	0.337	0.05	0.24	0.626	0.01	4.81	0.040*	0.19	0.46	0.506	0.02
group:direction:coordination	0.05	0.823	0.00	0.99	0.332	0.05	6.86	0.016*	0.26	9.54	0.006**	0.32

Significances are represented as following: * $p \leq 0.05$, ** $p \leq 0.01$, *** $p \leq 0.001$.

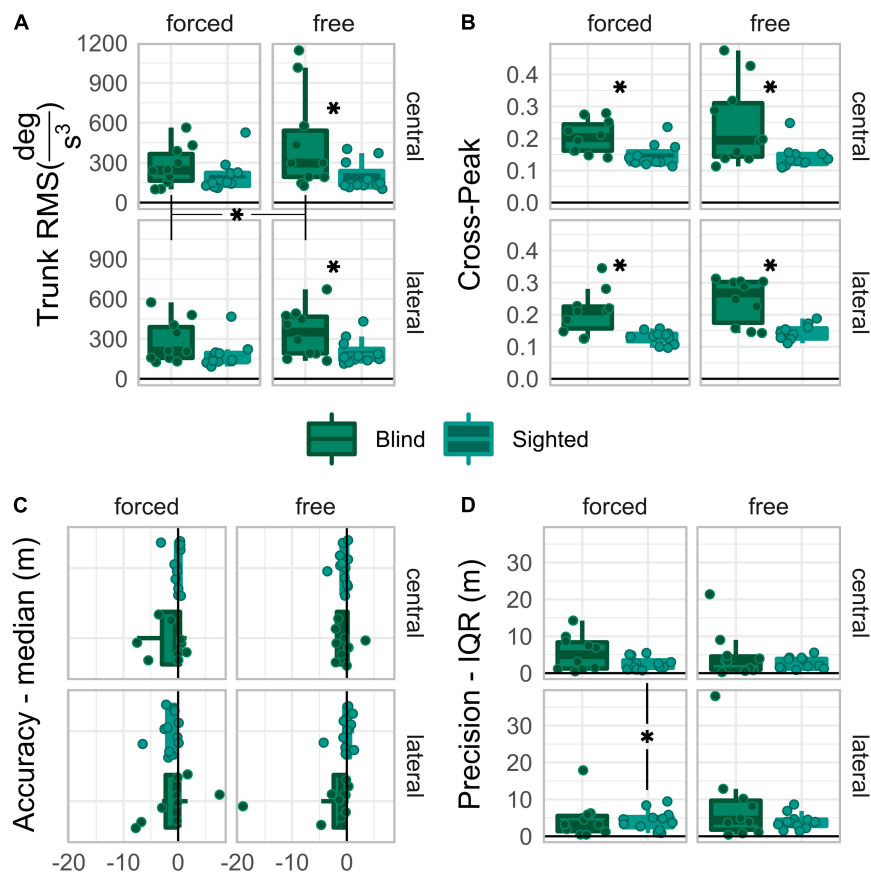


FIGURE 3 | Boxplots of trunk RMS (A), cross-peak (B), Accuracy (C), and Precision (D), reported for each group and driving condition. Asterisks expose the *post-hoc* comparisons significances. Those between graphs' labels expose the within-group main effects. Those in the space between subgraphs expose the 2-way within-group interaction effects. Those between boxplots in the same grid expose significant differences between groups.

central direction. The ART ANOVA test reached significance for group and control main effects, $F_{group}(1,19) = 5.23$, $p = 0.034$, $p.eta^2_{group} = 0.22$, $F_{control}(1,19) = 12.02$, $p = 0.003$, $p.eta^2_{control} = 0.39$, $F_{direction}(1,19) = 1.08$, $p = 0.312$, $p.eta^2_{direction} = 0.05$. Significance of the interaction effects was reached for “group:coordination,” $F(1,19) = 9.04$, $p = 0.007$, $p.eta^2 = 0.32$.

Post-hoc comparisons on the “group:coordination” interaction levels were performed under the alternative hypothesis that the

trunk jerk RMS is larger in blind than in sighted participants, with *free* rather than with *forced* coordination. **Figure 3C** shows that the trunk jerk RMS was significantly larger in blind than in sighted participants only when the control condition was *free*, $U_{blind-sighted|free} = 88$, $p = 0.02$, $r_{rb} = 0.60$, 95%CI [0.18,0.93], $U_{blind-sighted|forced} = 75$, $p = 0.173$, $r_{rb} = 0.36$, 95%CI [-0.15,0.84]. The RMS was significantly larger for the *free* than for the *forced* coordination level only in the blind group, $W_{free-forced|blind} = 54$, $p = 0.004$, $r_{rb} = 0.96$, 95%CI [0.64,1],

while the sighted group did not show any significant control-related change, $W_{\text{free-forced}|sighted} = 45$, $p = 0.32$, $r_{rb} = 0.36$, 95%CI $[-0.24, 1]$.

Cross-Peak

If early blind people could not immobilize their trunks, their cross-peaks would be higher than sighted participants, at least in the *forced* coordination level where sighted people are supposed to immobilize trunk movement. If having targets straight-ahead facilitated trunk immobilization, the cross-peaks would be larger in the *lateral* than in the *central* direction. The ART ANOVA test reached significance for group and control main effects only, $F_{\text{group}}(1,19) = 18.4$, $p < 0.001$, $p.\eta^2_{\text{group}} = 0.49$, $F_{\text{control}}(1,19) = 6.02$, $p = 0.024$, $p.\eta^2_{\text{control}} = 0.24$, $F_{\text{direction}}(1,19) = 0.05$, $p = 0.826$, $p.\eta^2_{\text{direction}} = 0.00$. No significance was reached in any interaction effect.

Even though no interaction effect reached significance, the “group:coordination” interaction approached significance and had a relatively large effect size, $F(1,19) = 3.59$, $p = 0.074$, $p.\eta^2 = 0.16$. Therefore, *post-hoc* comparisons were performed on the levels of the “group:coordination” interaction, under the alternative hypothesis, that the cross-correlation peaks are larger in blind than in sighted individuals, and for *free* than for *forced* levels. **Figure 3D** shows that the crosscorrelation peaks were significantly larger in blind than in sighted participants regardless of control, $U_{\text{blind-sighted}|free} = 98$, $p = 0.002$, $r_{rb} = 0.78$, 95%CI $[0.40, 1]$, $U_{\text{blind-sighted}|forced} = 96$, $p = 0.003$, $r_{rb} = 0.75$, 95%CI $[0.36, 0.96]$, and that no significant difference held between *free* and *forced* in any group, $W_{\text{free-forced}|blind} = 45$, $p = 0.084$, $r_{rb} = 0.64$, 95%CI $[-0.09, 1]$, $W_{\text{free-forced}|sighted} = 43$, $p = 0.413$, $r_{rb} = 0.30$, 95%CI $[-0.33, 0.94]$.

To summarize, sighted people had small trunk RMS, unaffected by the coordination factor. Contrarily, blind people had significantly larger trunk jerk RMS with *free* than with *forced* coordination; early blind people had significantly larger trunk jerk RMS than sighted people only with *free* coordination. Moreover, early blind people had higher cross-correlation peaks than sighted individuals in every condition.

Performance

The performance was assessed in terms of accuracy and precision. Accuracy was evaluated as the median of the by-trial final error. A negative value means a bias toward the hemifield of the initial direction. A positive value means bias away from the initial hemifield. Better accuracy means a value closer to zero. Precision was evaluated as IQR of the by-trial final error. Smaller IQR means better precision.

Accuracy

If the head-trunk coordination demand impaired participants' accuracy in steering toward the target, they would be less accurate with *forced* than with *free* coordination. If having targets straight-ahead facilitated localization, accuracy would have more negative values in *lateral* than in *central* direction. The ART ANOVA test reached significance for none of the main effects, $F_{\text{group}}(1,20) = 0.19$, $p = 0.665$, $p.\eta^2_{\text{group}} = 0.01$,

$F_{\text{direction}}(1,20) = 1.08$, $p = 0.312$, $p.\eta^2_{\text{direction}} = 0.05$, $F_{\text{control}}(1,20) = 0.02$, $p = 0.881$, $p.\eta^2_{\text{control}} = 0.00$. Significance of the interaction effects was reached for “group:coordination,” $F(1,20) = 6.87$, $p = 0.016$, $p.\eta^2 = 0.26$, and “group:direction:coordination,” $F(1,20) = 9.54$, $p = 0.006$, $p.\eta^2 = 0.32$.

Post-hoc comparisons on “group:direction:coordination” interaction levels were performed under the alternative hypothesis that accuracy is different between early blind and sighted, between *free* and *forced* levels, and more negative in *lateral* than in *central*. **Figure 3A** shows that none of the comparisons reached significance; however, the *central* vs. *lateral* comparison with *forced* coordination approached significance very closely in sighted, $W_{\text{lateral-central}|sighted:forced} = 12$, $p = 0.051$, $r_{rb} = -0.69$, 95%CI $[-1, -0.15]$, but not in blind individuals, $W_{\text{lateral-central}|blind:forced} = 23$, $p = 1.000$, $r_{rb} = -0.16$, 95%CI $[-0.85, 0.6]$.

Precision

If the head-trunk coordination demand impaired participants' precision in steering toward the target, they would be more precise with *free* than with *forced* coordination. If having targets straight-ahead facilitated localization, precision would be better in *central* than in *lateral* direction. The ART ANOVA test reached significance for none of the main effects, $F_{\text{group}}(1,20) = 0.39$, $p = 0.539$, $p.\eta^2_{\text{group}} = 0.02$, $F_{\text{direction}}(1,20) = 3.28$, $p = 0.085$, $p.\eta^2_{\text{direction}} = 0.14$, $F_{\text{control}}(1,20) = 0.9$, $p = 0.355$, $p.\eta^2_{\text{control}} = 0.04$. Significance was reached for the interaction effects “group:coordination,” $F(1,20) = 1.64$, $p = 0.214$, $p.\eta^2 = 0.08$, and “group:direction:coordination,” $F(1,20) = 6.86$, $p = 0.016$, $p.\eta^2 = 0.26$.

Post-hoc comparisons on the “group:direction:coordination” interaction levels were performed under the alternative hypothesis that precision is better in sighted than blind, in *central* than *lateral* direction, and with *free* than *forced* coordination. **Figure 3B** shows that sighted, not blind, were significantly more precise in the *central* than in the *lateral* direction when the control was *forced*, $W_{\text{lateral-central}|sighted:forced} = 67$, $p = 0.040$, $r_{rb} = 0.72$, 95%CI $[0.26, 1]$, $W_{\text{lateral-central}|blind:forced} = 22$, $p = 1.000$, $r_{rb} = -0.20$, 95%CI $[-0.89, 0.49]$.

To summarize, we could not find group-wise differences related to the head-trunk coordination demand. Moreover, only sighted people had better spatial performance when the target was straight-ahead than to the side, but only when trunk immobilization was demanded.

DISCUSSION

The goal of this study was to test the hypothesis that early visual loss impairs head-trunk coordination development for orienting movements in the horizontal plane. The investigation was performed by testing a group of early blind people on a head-pointing task with dynamic auditory stimuli delivered in AVR. In some trial blocks, a head-trunk coordination constraint was set implicitly and explicitly, inducing participants to immobilize their trunks. Kinematic behavior and spatial performance were

assessed within-group by comparing trial blocks with and without coordination demand and between-group by comparing early blind participants with sighted blindfolded controls. The assessed aspects of kinematic behavior were similarity between head and trunk movements via cross-peak (head-trunk jerk cross-correlation peak amplitude) and trunk movement via trunk jerk RMS. The spatial performance was assessed by accuracy and precision: median and interquartile range of the distribution of the trial-by-trial final distances from the target center (**Figure 3**).

Two possible scenarios may have emerged. If early blindness did not impair head-trunk coordination for goal-directed horizontal rotation, no difference would have emerged in kinematic behavior or spatial performance. Alternatively, if early blindness impaired head-trunk coordination, early blind people would differ from sighted controls when demanded to coordinate head and trunk; this would definitely occur in their kinematic behavior and, if the impairment is large enough, also in spatial performance. Our results confirmed the hypothesis that early visual deprivation hampers the development of head-trunk coordination for orienting movements in the horizontal plane. Indeed, the early blind group showed an overall larger cross-peak than the blindfolded sighted group (**Figure 3B**). Moreover, when the trunk was free to move (*free* coordination), early blind people had larger trunk jerk RMS than sighted blindfolded in the same condition, and larger trunk jerk RMS when voluntary trunk immobilization was demanded (*forced* coordination) (**Figure 3A**). Altogether, the results concerning kinematic behavior describe the following scenario: early blind people, when free to rotate around the vertical axis, did so with head and trunk together; furthermore, when asked to immobilize their trunk, they did it by damping their trunk rotations (doing rotations with small amplitude) instead of avoiding them. Although the motor behavior exposed by the early blind group, hereafter named “damping” behavior, exposes some degree of head-trunk coordination, their head and trunk movements were more similar than those of sighted participants. The early blind group’s motor behavior recalled the “*en-bloc*” motor coordination strategy, typical of 3 to 8 year-old sighted children with incomplete head-trunk coordination development (Assaiante et al., 2005).

Several pieces of evidence, mainly from postural balance tasks (Easton et al., 1998; Schmid et al., 2007; Alotaibi et al., 2016), have suggested the existence of a link between early visual loss and head-trunk coordination deficit. Yet, to the best of our knowledge, this is the first study that directly identifies the connection for goal-directed orienting movements in the horizontal plane. The information obtained here is limited to acknowledging the existence of such a deficit; conclusions cannot be drawn about its etiology. Vision may be necessary for the development of head-trunk coordination, or it could act as the catalyst of a process driven, instead, by motor experience (Lopes et al., 2011). Indeed, visual disabilities are very often a great barrier for people to move freely (Marron and Bailey, 1982; Guralnik et al., 1994; West et al., 2002), and consequently limit their chances to explore and learn new motor commands, even the coordination of head and trunk.

One may expect a deficit in head-trunk coordination to have some consequences on spatial performance, at least when tasks require good motor control skills, i.e., the trial blocks with *forced* coordination. Our behavioral measures failed to identify differences between early blind and sighted groups (**Figures 3C,D**). This failure probably arises from the method’s inadequacy in causing the deficit to affect spatial performance: the implicit coordination demand was designed to affect performance if people could not immobilize their trunk. Early blind participants, instead, using “damping” behavior, managed to do so well enough that their performance was not affected. The “damping” behavior identified in this paper satisfactorily explains why early blind people perform as good as, or even better than sighted individuals, in horizontal sound localizations by head pointing (Lessard et al., 1998; Röder et al., 1999; Collignon et al., 2009; Lewald, 2013). Since generic head-pointing tasks with no or passive-only coordination constraints challenge head-trunk coordination less than our task, early blind people’s “damping” behavior compensates their head-trunk coordination deficit well enough not to let it affect head-pointing performance. It remains unclear whether the coordination deficit identified here can still cause performance drops in the unstructured setups found in everyday life, where motor and coordination demands are more complex and varied.

The comparison between trial blocks with targets in front of participants (*central*) versus those with targets to the side (*lateral*) provides another point of discussion in light of the literature on egocentric auditory localization. Past studies on the typical population have highlighted, using different methods, some kind of spatial performance drop when targets were placed at eccentric positions concerning the participants’ straight-ahead and when the head was turned (Makous and Middlebrooks, 1990; Lewald et al., 2000; Occhigrossi et al., 2021). Most of these studies identified a stimulus eccentricity underestimation bias, that is, accuracy loss. It was also shown that early blind people did not exhibit the underestimation bias and, therefore, obtained more accurate results than sighted people (Lessard et al., 1998; Zwiers et al., 2001; Lewald, 2013). Our results contribute to such body of evidence by identifying a precision, not accuracy, drop in sighted participants, only with *forced* coordination. At first glance, our results may appear in contrast with the previously identified accuracy drop in head-pointing localization. They complement the previous findings by showing what happens in a less structured context. The bias identified in head-pointing tasks was shown to be a function of head-on-trunk eccentricity (Makous and Middlebrooks, 1990; Lewald et al., 2000; Occhigrossi et al., 2021). Instead, in our task, the relative position between participant’s seat and sound source is continuously updated (participants “sit” on the virtual arrow, which advances in the virtual space at fixed speed). By doing so, each trial ends with a different head eccentricity. In our case, it is likely that the aggregation of trials that contain different head eccentricities, hence different biases, resulted in more dispersed samples. In support of this view is the fact that the pattern of our results on precision strictly matches the previous literature for accuracy when the coordination is *forced*, that is

when the trunk is voluntarily immobilized and head-on-trunk rotations are maximized.

To conclude, early visual deprivation affects the full development of head-trunk coordination for orienting movements in the horizontal plane, yet the degree of control over the trunk obtained without early visual experience is enough to dampen unwanted trunk rotations. This “damping” strategy lets early blind people perform head-pointing tasks unaffected, even when sounds are not static and coordination constraints are demanded. The etiology of this deficit remains unclear and will be the object of further investigation, as well as the impact of the coordination deficit on the performance of more complex tasks such as steering during locomotion or reaching to targets placed sideways. Future experimental paradigms shall more closely reflect daily life activities, such as shopping at the grocery store (as an example study, see Kim et al., 2020).

DATA AVAILABILITY STATEMENT

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found below: <http://doi.org/10.5281/zenodo.4707477>.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Comitato etico, ASL 3, Genova. The

patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

DE developed the virtual reality platform and collected and analyzed the data. All authors designed the experiment, wrote and approved the manuscript, contributed to the article, and approved the submitted version.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: <https://www.frontiersin.org/articles/10.3389/fnhum.2021.699312/full#supplementary-material>

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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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fNIRS Monitoring of Infant Prefrontal Cortex During Crawling and an Executive Functioning Task

Hannah Weibley, Mina Di Filippo, Xinran Liu, Lillian Lazenby, Jackson Goscha, Alyssa Ferreira, Laura Muscalu and Nancy Rader*

Psychology Department, Ithaca College, Ithaca, NY, United States

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Silvio Ionta,
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Marieke R. Gilmartin,
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Imaging and Bioengineering (NIBIB),
United States

*Correspondence:

Nancy Rader
rader@ithaca.edu

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Functional near-infrared spectroscopy (fNIRS) is a brain-imaging technology used to reveal brain activity by measuring blood oxygenation. Using fNIRS we measured activity in the left prefrontal lobe of 8–14 month-old infants as they crawled or were pushed in a stroller and as they were given a passive attention task or an active executive function (EF) task. For each task, we measured peak total hemoglobin concentration and peak Oxy relative to baseline. Results revealed differences in peak Oxy levels for crawling vs. strolling and between the EF and passive cognitive tasks, with total hemoglobin greater for the EF task than the passive attention task. These results support the theoretical view that both active locomotion and EF engage the prefrontal cortex (PFC) during early development.

Keywords: fNIRS, executive function, prefrontal cortex, locomotion, attention, infancy

INTRODUCTION

In both children and adults, the prefrontal cortex (PFC) is an area linked to executive functioning, a set of neurocognitive and regulatory processes. There are three main components to executive functions (EF): inhibitory control, cognitive flexibility, and working memory. Each function plays a role during situations that require controlled processing and selective attention. Executive functions allow us to self-regulate, task switch, problem-solve, and are overall essential to learning. They can also serve as a predictor in childhood for later cognitive development and quality of life (Diamond, 2013). However, very little is known about the development of executive function in infancy.

Koziol et al. (2011) have proposed that executive function processes in the human brain developed over evolution through pressures derived from the need to control motor behavior. This perspective links self-guided locomotion and the development of the executive function, with both functions making use of the prefrontal lobe of the brain (e.g., Koziol and Lutz, 2013). While Koziol et al. (2011) claim that locomotion and executive function share brain areas, there is no research that directly evaluates their claim for this sharing in human infants. Indirect support for this view was found in research with non-crawling infants who began at 5 months of age using a robotic locomotive device. After 2 months infants who locomoted with the robotic device performed better on executive function tasks at 7 months compared with infants who did not have that locomotive experience (Rader et al., 2019). Given the relationship reported between motor impairments and delays with poor executive function (Leonard, 2016),

Kozioł et al.'s (2011) theory has important clinical implications as well as serving as a potential key to understanding the development of executive function in a typically developing population. The key aim of the reported research was to explore, using functional near-infrared spectroscopy (fNIRS), the prefrontal lobe as a shared brain area for intentional locomotion and executive function in infants.

The cortical activity accompanying executive functions can be measured with technology such as fMRI and fNIRS. These technologies provide data that reflect the brain's responsiveness to stimuli or processes needed for performing particular actions. fNIRS technology has become an increasingly popular neuroimaging technique, particularly within pediatric populations (Aslin, 2012), as it is safe and allows for subject movement. León-Carrión and León-Domínguez (2012) verified fNIRS as an accepted neuroimaging technique that could be applied in both clinical and research settings.

When neurons are activated, their metabolic demands change, resulting in an increase in oxygen consumption, local cerebral blood flow, and oxygen delivery (Lloyd-Fox et al., 2010). According to Villringer and Chance (1997), a typical hemodynamic response to neurons being activated in the prefrontal cortex (PFC) is an increase in blood flow, generating an increase in oxygenated hemoglobin (oxy-Hb), as well as a slight decrease in deoxygenated hemoglobin (deoxy-Hb) and an overall increase in total hemoglobin (HbT). In research with adults, it has been established that fNIRS can be used to measure cognitive workload (Ayaz et al., 2012) and task difficulty (Izzetoglu et al., 2004) by measuring changes in oxy-Hb, deoxy-Hb, and HbT in the prefrontal cortex (Herff et al., 2014). Also, it has been demonstrated that increased cognitive demands result in increased activation of Brodmann Area 10 (BA10), located in the prefrontal cortex (Arsalidou et al., 2013). Wager et al. (2004) associate BA10 with executive functioning and Wager et al. (2005) describe BA10, specifically the left anterior PFC, as active during tasks that require the inhibition of learned rules/behavior.

Researchers have also used fNIRS to measure activation in the prefrontal cortex during motor activities in adults. For example, Suzuki et al. (2004) investigated prefrontal cortex activation while participants walked and ran on a treadmill and found that the prefrontal cortex, as well as the premotor cortex, showed activation when adjusting to accelerating speeds on the treadmill. Lloyd-Fox et al. (2008) recorded prefrontal activity during external perturbation trials, which revealed that the prefrontal cortex plays an important role in balance control. Using a clinical population, Maidan and colleagues (e.g., Maidan et al., 2015, 2016, 2017) have reported a connection between BA10 and locomotor control in Parkinson's Disease patients.

Two issues arise when recording from a single brain area. One is that it is not possible to know the contribution of other areas of the brain. In the research reported here, the goal has been to examine the extent to which the prefrontal cortex is activated during goal-directed locomotion and executive functioning without analyzing the contribution of other brain areas. Aslin (2012) has raised a related concern; Aslin questions the extent to which any increase in brain activity in a particular

region might be explained by its role as a general index of attention or arousal. Laeng et al. (2012) have presented a case for using pupil dilation to track arousal since it is a response created by norepinephrine produced by the locus coeruleus, a structure in the brainstem that regulates the integration of the brain's attention system, stress response, and arousal regulation. Rader and Zukow-Goldring (2015) have successfully used pupil dilation with infants as a measure of arousal. That activity in the prefrontal cortex as measured using fNIRS and pupil dilation can be separated is shown by YTS Brighter and Rader (2018) who found different patterns in pupil diameter and oxygenation response in the participants' reactions to editing shifts in video segments, suggesting differences between pupil diameter and fNIRS in terms of the psychological phenomena they measure.

In this study, we used fNIRS to record activity from the prefrontal cortex of crawling infants during active, goal-directed, locomotion, where they crawled to reach a parent, and passive locomotion, where they were pushed in a stroller to reach a parent. The prefrontal activity was also measured during two cognitive tasks, one that involved passive attention to a dancing cat puppet and the other that involved executive function. For the executive function task, known as a "switch task," participants learned a rule and then had to inhibit it and switch to a new rule (Kovács and Mehler, 2009). Additionally, we measured pupil diameter during the cognitive tasks to provide an index of arousal.

We hypothesized that there would be higher peak Oxy (the difference between oxy-Hb and deoxy-Hb) and HbT levels relative to baseline when infants actively crawled compared to being passively moved in a stroller. We also predicted that these oxygenation measures would be higher, compared with baseline, during the Switch Task as compared with the Cat Puppet Task. Additionally, we set out to determine the relationship between the fNIRS effects and pupil diameter.

MATERIALS AND METHODS

Participants

Inclusion criteria for this study required participants to be within the age range of 8–15 months old, typically developing, crawling at the time of participation, able to tolerate wearing the apparatus, and able to participate in both sessions of the study. Of the 20 infants who met these criteria and came to the lab, data from eight infants could not be analyzed either because of infant fussiness that occurred during testing (five infants) or because of experimenter error (three infants).

The 12 infants whose data are described here were six males and six females, aged 8.3–15.4 months, with an average age of 11.1 months ($SD = 2.17$). Ten infants were identified as Caucasian and two as an ethnic minority, reflective of the local population.

Parents of the participants gave informed consent prior to their child's participation in the study. Parents received a \$20 gift card following each of the two sessions, a certificate of completion, and a photo for their time and contribution to the research. Most participants were recruited through the campus



FIGURE 1 | Pediatric sensor strip and infants in the crawling and stroller conditions.

online newsletter, use of local parental pages, and other social media platforms. Approval for this research was received from the Ithaca College Institutional Review Board.

Design and Apparatus

This study used a within-subjects design to collect data on the 12 infant participants. The experiment was carried out in two separate sessions, completed no more than 2 weeks apart. Session 1 provided measures of oxygenation levels in the brain during crawling and stroller movement, while Session 2 provided measures of oxygenation levels in the brain during a passive attention task and during an active cognitive task requiring executive function.

Functional Near-Infrared Spectroscopy

fNIRS technology uses LED light and optical sensors to measure the concentration changes from baseline in oxygenated hemoglobin (oxy-Hb) and deoxygenated hemoglobin (deoxy-Hb) in the capillaries (León-Carrión and León-Domínguez, 2012). Blood oxygenation can be differentiated due to oxy-Hb and deoxy-Hb having different absorption properties to near-infrared light exposure and varying light scattering patterns when the light is reflected back from the brain (Lloyd-Fox et al., 2010).

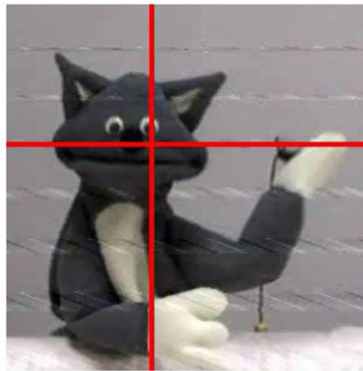
Our fNIRS device (Biopac Systems, 2018) emits infrared light having wavelengths of 730 nm and 850 nm, under which biological tissue essentially becomes translucent (Lloyd-Fox et al., 2010). We used a Biopac pediatric sensor (RXFNIR-PED) to measure light levels reflected back from the oxygenated and deoxygenated hemoglobin; this sensor has two channels, two detectors, and one emitter, with an inter-optode

distance of 20 mm (see **Figure 1**). The sensor is sized for infants to record from a single hemisphere of the prefrontal cortex.

Oxy-Hb and deoxy-Hb levels were measured within a light level of less than 4,000 mv. To operate within this range, the LED current (brightness) and detector gain (sensitivity) were adjusted for each participant as determined by CobiStudio, software distributed by Biopac (Ayaz, 2005); typical brightness (LED current) was 12 mA and a gain of 2. Because we were working with infants, a sample rate of 50 per optode per frame was used. Using CobiStudio, event markers were programmed manually to signify the beginning and end of specific tasks within the study. Markers were also used to indicate any intervals in which the infant was briefly fussing or crying, and these intervals were excluded from analyses. Infants who fussed or cried repeatedly during a trial are not included in our sample.

Each data output file was refined using a low pass filter, which was created for the pediatric wireless device, and a motion artifact rejection algorithm. The low pass filter was set at 0.1 Hz with an order of 50, Hamming. The motion artifact rejection filter [Sliding-Window Motion Artifact Rejection (SMAR)] eliminates any data that are associated with light oversaturation and other abnormalities caused by the movement of the sensor on the skin. Details of these filters and the Modified Beer Lambert Law used can be found in Ayaz (2010) and Ayaz et al. (2010). Final analyses of the infrared light and oxygenation data were conducted using fNIRSoft software.

We recorded hemodynamic responses from the left anterior prefrontal cortex. While it cannot be definitively determined for infants, we believe we measured responses from BA 10,



Switch Task Components: Fixation, Anticipation, Puppet

FIGURE 2 | The cat puppet and switch task.

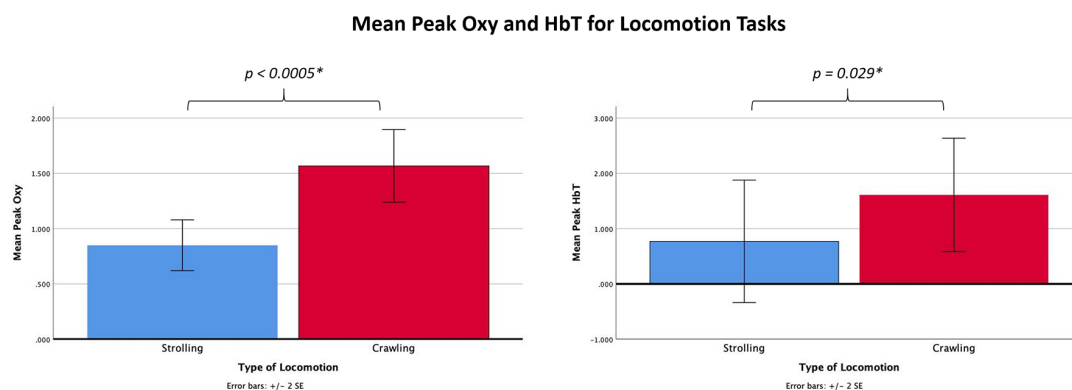


FIGURE 3 | Graphs showing the mean peak Oxy and HbT for the locomotion tasks.

comprising the most anterior part of the prefrontal cortex. The fNIRS pediatric sensor was placed over the left side of the infant's forehead, centered just above the pupil. The sensor was held in place by a black headband and then covered with a cotton hat (see **Figure 1**) to secure the sensor strip and to prevent light leakage. The sensor information was transmitted to a computer *via* a wireless interface secured behind the infant's back in the zippered pouch of a custom-made vest.

The hemodynamic measures used as dependent variables in this research were Peak Oxy and Peak HbT. A peak indicates the maximum value obtained during a particular task. The peak response was selected as a dependent measure value because the response in infants over time is unknown and variable. Using a peak measure allows a fair comparison across infants and also across tasks. Oxy levels reflect changes in oxygenation concentration determined by the difference between oxy-Hb and deoxy-Hb relative to baseline. HbT levels indicate the total hemoglobin concentration, relative to baseline, and reflect changes in blood volume. The final 3 s of baseline was used in all cases for the data analyses.

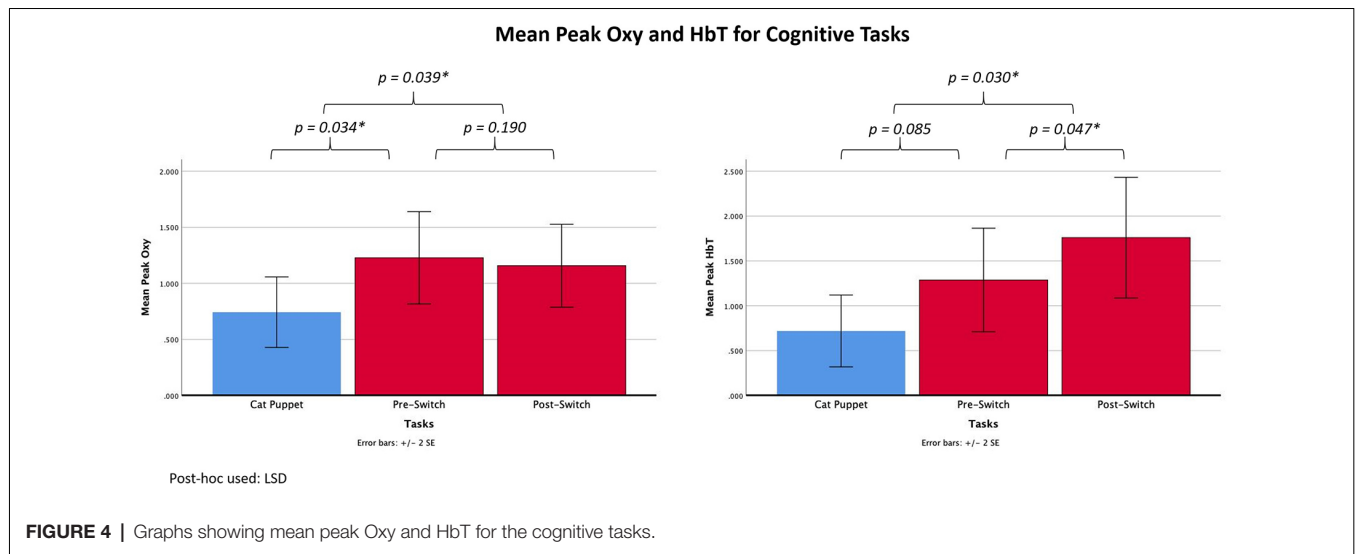
Procedure

Session 1

The focus of the study's first session was locomotion. After the fNIRS device was placed on the participant's forehead with the emitter placed directly over the pupil of the left eye, they were brought into the testing room for the locomotion trials. For the passive movement portion, the participant was pushed in a stroller for a distance of 12 feet towards their parent who called to them and/or enticed them with a toy (see **Figure 2**). For the active movement portion, the participant crawled 12 feet towards their parent who encouraged them in a fashion similar to the stroller condition. The fNIRS baselines lasted 15 s and were established during the time that the participant was stationary prior to locomotion. The order of crawling and stroller events was counterbalanced across participants. We allowed for two trials each of crawling and strolling with the intention of having one trial without recording issues.

Session 2

The focus of the second session was on responses to cognitive tasks. Participants were placed in a car seat in a testing room with a large plasma screen on which video segments were shown.



An eyetracking system (Applied Science Laboratory) was used to track infant eye movements in relation to the video scenes so that it could be determined in real time that the infant was paying attention to the video being presented. A brief clip from a show called *Teletubbies* was shown initially to obtain the participant's attention, establish a baseline of oxygenation levels in the prefrontal cortex, and allow experimenters to capture the participant's eye for eyetracking.

Following *Teletubbies*, two different cognitive-visual tasks occurred. The first task, the Cat Puppet Task, displayed a cat puppet that danced to music for 15 s. This task reflects passive attention to a stimulus. The second task, the Switch Task, reflects active attention, requiring infants to learn a rule and then inhibit the learned rule and switch to a new rule. Kovács and Mehler (2009) present this task as a type of executive function task. At the start of the task, a puppet appears on the right side of the screen; this occurs for nine consecutive trials (Pre-Switch). After the ninth trial, the puppet switches to the left side of the screen, appearing for another nine consecutive trials (Post-Switch). At the beginning of each trial, a visual cue appears in the center of the screen to capture the attention of the infant and bring the focus back to the center of the screen. The total duration for each Switch Task component was 54 s (see images for both Tasks in Figure 2).

RESULTS

Comparisons between crawling and stroller locomotion were performed on data from 11 of the 12 participants, as one infant had difficulty completing the crawling task. Two paired-samples *t*-tests were conducted to compare peak Oxy and peak HbT levels (the dependent variables) between crawling and stroller trials (levels of the independent variable). Significant differences between the stroller and crawling tasks were found for both peak Oxy, $t_{(10)} = 6.85$, $p < 0.005$, $\eta^2 = 0.824$, and peak HbT, $t_{(10)} = 2.55$, $p = 0.029$, $\eta^2 = 0.395$. For peak Oxy, means (standard deviations) were 0.85 (0.38) for strolling and 1.57 (0.55) for

crawling. For peak HbT means (standard deviations) were 0.77 (1.84) for strolling and 1.61 (1.70) for crawling. These results are shown in Figure 3.

Two repeated-measures ANOVAs with one independent variable (Task) were performed to analyze peak Oxy and peak HbT (the dependent variables) during the three cognitive tasks (the Cat Puppet, Pre-Switch, and Post-Switch). There was an overall Task effect for Oxy, $F_{(2,22)} = 3.836$, $p = 0.037$, $\eta^2 = 0.259$. Subsequent paired comparisons using LSD showed that the values for the Cat Puppet Task differed significantly from the Pre-Switch ($p = 0.034$) and Post-Switch ($p = 0.039$) Tasks; the Pre-Switch Task did not differ significantly from Post-Switch ($p = 0.717$). Means (standard deviations) were 0.7427 (0.5451) for the Cat Puppet Task, 1.228 (0.7126) for Pre-Switch, and 1.1572 (0.6403) for Post-Switch, as shown in Figure 4. There was also an overall task effect for HbT, $F_{(2,22)} = 5.282$, $p = 0.013$, $\eta^2 = 0.324$. Paired comparisons using LSD showed that the Cat Puppet Task differed significantly from Post-Switch ($p = 0.039$) but did not reach a value of $p < 0.05$ in comparison with Pre-Switch ($p = 0.085$). Post-Switch values differed significantly from Pre-Switch ($p = 0.047$) values. Means (standard deviations) were 0.7189 (0.6938) for the Cat Puppet Task, 1.2871 (1.000) for Pre-Switch, and 1.759 (1.1663) for Post-Switch, as shown in Figure 4.

We performed correlation analyses to look at the relationship of pupil diameter to the hemoglobin measures during the cognitive tasks. The pupil diameter values were provided by Gazetracker software by Eye Response Technologies and acquired as part of Applied Science Laboratory's eye tracking system. No statistically significant correlations were found between average pupil diameter and peak Oxy or between pupil diameter and peak HbT. For the Cat Puppet Task, the *r*-value for Oxy was 0.26 and for HbT 0.22; for Pre-Switch the *r*-values were 0.56 and 0.55, while for Post-Switch they were 0.19 and 0.31. Of interest is the fact that the pupil diameter values for the Cat Puppet Task and both Switch Tasks were significantly correlated—Cat Puppet with Pre-Switch ($r = 0.79$, $p < 0.01$)

and with Post-Switch ($r = 0.61$, $p < 0.05$) and Pre-Switch with Post-Switch ($r = 0.77$, $p < 0.01$).

DISCUSSION

The goal of this study was to examine the link of the prefrontal cortex to both self-guided, goal-directed locomotion and executive functioning during infancy. The data support our hypothesis that there would be higher Oxy levels when infants actively crawled compared to when they were passively moved in a stroller. Additionally, the data support the prediction of higher Oxy levels during the Switch Task as compared with the Cat Puppet Task. Moreover, based on our data, the differences in peak HbT levels between the Cat Puppet Task and the Switch Task suggest that more cognitively demanding tasks enlist a higher blood volume.

The findings of our study support Koziol and Lutz's (2013) hypothesis that both active locomotion and executive function tasks engage the prefrontal cortical area of the infant brain. In a goal-oriented activity like crawling to a parent, one cannot separate the effect of the motor activity from the controlling of that activity. According to Koziol and Lutz's (2013) theory, the need to control body movements for effective locomotion in infancy has recruited a brain area within the prefrontal cortex, namely BA 10, that has subsequently become the locus of more general executive function control. Indeed, our results show that an activity that requires mental coordination and inhibition of an established behavior (the Switch Task) and an activity that required motor coordination and control (goal-directed crawling) activate more strongly an area of the prefrontal cortex than passive activities (being pushed in a stroller or viewing the Cat Puppet dancing). This finding has implications for understanding the reported developmental relationship between delayed or impaired motor function and cognitive skills (Leonard, 2016).

It could be argued that in the present study the hemodynamic patterns observed during crawling and during the Switch Task reflect a higher level of attention/arousal rather than executive control, unnecessary during the Cat Puppet Task. However, the high correlations for pupil diameter across the three cognitive tasks suggest that the differences across these tasks in the hemodynamic responses cannot be completely due to differences in arousal/attention.

While not uncommon in fNIRS research, a limitation of this study is its small sample size and limited representation of ethnic groups. Also, we measured activity only in the left hemisphere because we found during pilot testing that infants were unable to tolerate the doubling of time required to measure from both hemispheres. The need for the doubled time occurs because

the pediatric sensor allows for measurements from only one hemisphere. One rationale for using the left hemisphere rather than the right hemisphere is Wager et al.'s (2005) finding with adults that the left anterior PFC is active during tasks that require the inhibition of learned rules/behavior.

With these limitations in mind, we have found, for crawling-aged infants, that the left anterior prefrontal cortex is more likely to be engaged during both active locomotion and a cognitive task requiring executive function than when passively moved through the same environment or when watching a video requiring only passive attention. Future research might be directed towards looking at the hemodynamic responses in other areas of the brain to get a more complete picture of the complexities of the development of executive function in infants.

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 Institutional Review Board Ithaca College. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin. Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

Suzanne Zuckerman began this research as her senior honors project at Ithaca College, troubleshooting the fNIRS set-up, developing the testing protocol, and conducting pilot research. The first six authors (HW, MD, XL, LL, JG, and AF) participated in testing infants and in contributing to the manuscript for this article. In addition, HW and MD acquired and applied their knowledge of fNIRS principles and along with XL, coded the data and carried out the statistical analyses. MD acquired the pupil diameter data and carried out the pupil diameter analysis. LM and NR supervised all aspects of the conduction of this research. All authors contributed to the article and approved the submitted version.

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The Age-Related Changes in Speed of Visual Perception, Visual Verbal and Visuomotor Performance, and Nonverbal Intelligence During Early School Years

Rana J. Alghamdi^{1,2*}, Melanie J. Murphy¹, Nahal Goharpey¹ and Sheila G. Crewther¹

¹Department of Psychology and Counselling, La Trobe University, Melbourne, VIC, Australia, ²Department of Psychology, King Saud University, Riyadh, Saudi Arabia

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*Correspondence:

Rana J. Alghamdi
18396777@students.latrobe.edu.au

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Speed of sensory information processing has long been recognized as an important characteristic of global intelligence, though few studies have concurrently investigated the contribution of different types of information processing to nonverbal IQ in children, nor looked at whether chronological age vs. months of early schooling plays a larger role. Thus, this study investigated the speed of visual information processing in three tasks including a simple visual inspection time (IT) task, a visual-verbal processing task using Rapid Automatic Naming (RAN) of objects as an accepted preschool predictor of reading, and a visuomotor processing task using a game-like iPad application, (the “SLURP” task) that requires writing like skills, in association with nonverbal IQ (Raven’s Coloured Progressive Matrices) in children ($n = 100$) aged 5–7 years old. Our results indicate that the rate and accuracy of information processing for all three tasks develop with age, but that only RAN and SLURP rates show significant improvement with years of schooling. RAN and SLURP also correlated significantly with nonverbal IQ scores, but not with IT. Regression analyses demonstrate that months of formal schooling provide additional contributions to the speed of dual-task visual-verbal (RAN) and visuomotor performance and Raven’s scores supporting the domain-specific hypothesis of processing speed development for specific skills as they contribute to global measures such as nonverbal IQ. Finally, RAN and SLURP are likely to be useful measures for the early identification of young children with lower intelligence and potentially poor reading.

Keywords: sensory processing speed (PS), young school-age children, visual inspection time, visual verbal RAN processing, SLURP visuomotor processing, Raven’s Nonverbal intelligences

INTRODUCTION

Significant correlations between measures of speed of information-processing (inspection time and reaction time tasks) and intelligence were first described in young adults more than 40 years ago by Vickers et al. (1972) and others later on (Jensen and Munro, 1979; Vernon, 1983; Nettelbeck et al., 1986; Deary et al., 2001; Grudnik and Kranzler, 2001; Jensen, 2006, 2011; Sheppard and Vernon, 2008). Around this time brain imaging techniques were

appearing and Positron Emission Tomography (PET) was used to demonstrate that smarter adult brains work more efficiently with faster rates of information processing and utilize less neural energy than individuals with lower IQ scores (Haier et al., 1988, 1992; Jung and Haier, 2007).

Since then the speed of visual information processing has been investigated using a variety of paradigms including simple sensory perceptual tasks such as visual inspection time tasks, more complex coding tasks, and dual response time tasks (Blake, 1974; Kail and Park, 1992; Miller and Vernon, 1997; Weiler et al., 2003; McAuley and White, 2011). In particular, processing speed in children has been shown to increase with chronological age (Case, 1985; Anderson, 1996; Anderson et al., 1997). Indeed, in a review of 72 published studies, Kail (1991a) found that motor reaction times of young children (4–5 years) to visual stimuli were a third of the rate of adults, whereas older children (8 years) performed only twice as slowly as adults, raising the questions of whether: (i) these age-based changes in processing time were predominantly cognitive or motor-based development; (ii) whether the improved speed was more aligned to chronological age or domain-specific school routine based changes (Chi, 1977; Logan, 1988), and (iii) to what extent chronological age or months attending formal schooling (MAS), the related increase in the rate of visual processing mediates non-verbal IQ.

Thus, this study aimed to investigate rates of visual processing and whether differences in visual cognitive processing speed were due to individual school type experiences learning to read and write (domain-specific knowledge) or a single, global mechanism such as fluid nonverbal IQ that drives the exponential rate of visual information processing speed during child development (Kail and Park, 1992). To do this, we concurrently assessed the contribution of rates of simple, non-motor measured visual object recognition (Inspection Time task) modified from Vickers et al. (1972), visuo-verbal information processing (Rapid Automatic Naming of objects), and visuomotor eye-hand co-ordination and age and schooling to the prediction of non-verbal IQ (a global mechanism). The RAN of familiar objects task was chosen both as a measure of visual object verbalization and because it is a well-accepted predictor of future reading ability (Denckla and Rudel, 1974; Crewther et al., 2011, 2017; Siddaiah and Padakannaya, 2015; Savage et al., 2018; Landerl et al., 2019; Peters et al., 2020). Visuomotor skills have traditionally been assessed in terms of complex tasks with emphasis on manual timing (Tiffin and Asher, 1948; Wilson et al., 2000; Hart et al., 2006), rather than as measures of speed of accurate eye-hand coordination. Hence, the current

study assessed visuomotor performance by sensitively recording the time taken and errors made in tracing five prescribed shapes in an iPad app known as the SLURP task (SLURP; Lee et al., 2014). Age-related contributions to nonverbal IQ were also measured using the raw scores on the Raven's Colour Progressive Matrices (RCPM) nonverbal measure of reasoning ability, rather than standard scores that are corrected for developmental changes (Fry and Hale, 2000) and hence, likely to confound investigations of the age-related differences in multiple age groups. We expected that months of formal schooling would independently contribute to the development of the visual-verbal processing and visuomotor skills required for early school year foci of reading and writing (Burrage et al., 2008; Brod et al., 2017; Morrison et al., 2019), rather than simple visual perception, in line with the domain-specific knowledge hypothesis.

MATERIALS AND METHODS

Participants

One hundred primary school beginners (Males = 49 and Females = 51; Prep $n = 57$, Grade 1 $n = 28$, and Grade 2 $n = 15$), and three age groups; 5 years ($n = 31$), 6 years ($n = 39$) and 7 years ($n = 30$) were recruited from three primary schools in metropolitan Melbourne, Australia (see **Table 1**). Parents/guardians received a written description of the research tasks and were informed that they could withdraw their child from the study at any stage as per the Declaration of Helsinki. Parents/guardians provided written consent for their child to take part in the study and verbal consent to participate in the study was also obtained from children prior to the commencement of testing sessions. This study was conducted with approval from the La Trobe University Human Ethics Committee, the Victorian Department of Education Human Ethics Committee, and the Victorian Catholic Schools Ethics Committee (HEC 18139). Inclusion criteria required adequate vision and hearing, neurotypical development, and age appropriate English-speaking ability.

Materials

Screening Measures

Nonverbal Intelligence

Nonverbal IQ was measured using the (Raven's, 1958, 1995) Coloured Progressive Matrices (RCPM) a well-normed culture, and language-free psychometric test of non-verbal reasoning (Raven et al., 1998; Cotton et al., 2005). The RCPM consists

TABLE 1 | Mean (SD) and range for chronological age (years), Grade and nonverbal IQ (RCPM) raw scores for each age group.

Age group	N	M (SD)	IQ	(SD)	Range
5 years old	31	5.62 (0.22)	18.37	3.64	13–18
6 years old	39	6.38 (0.28)	20.54	4.54	12–30
7 years old	30	7.48 (0.33)	26.37	3.88	17–32
Prep	57	5.91 (0.38)	19.18	4.08	12–30
Grade 1	28	6.81 (0.40)	24.89	4.53	17–32
Grade 2	15	7.68 (0.26)	26.50	3.98	20–32

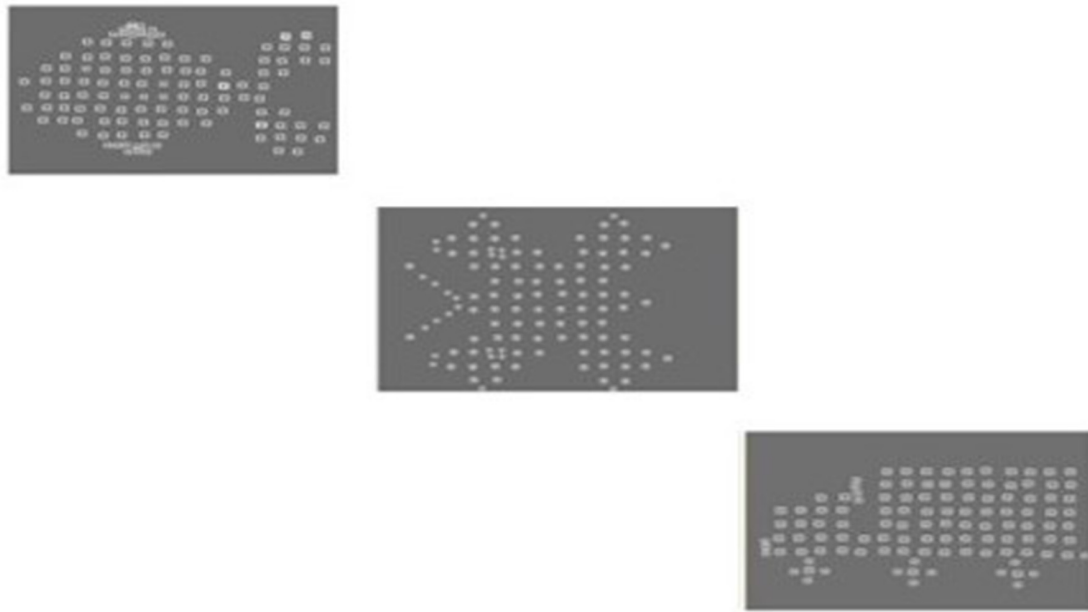


FIGURE 1 | The fish, butterfly, truck (FBT) Inspection Time task consisting of three images rapidly and randomly presented.

of 36 colored matrices. Each matrix has one piece missing, and participants are asked to choose the most appropriate missing piece from six possible options. This task was developed for individuals between 5–11 years old and presents with high-reliability $r = 0.80$ (Raven et al., 1998; Cotton et al., 2005).

Experimental Measures

Inspection Time (IT) as a Simple Measure of Visual Object Recognition

A non-motor IT task modified from Vickers et al. (1972) by Brown and Crewther (2017) and Ebaid and Crewther (2019) was used to assess visual information processing and visual attention. This task is a simple computerized measure of the minimum time required to identify one of three simple stimuli flashing on the screen (Fish, Truck, or Butterfly) using a PEST (parametric estimation of statistical threshold) routine (see **Figure 1**). Following the presentation, participants verbally indicated which one of the three images they saw—A Fish, Truck, or Butterfly and the examiner clicked the corresponding keyboard arrow button (Fish = ←, Truck = ↓, Butterfly = →). Each child completed 32 trials which took approximately 5 min in duration. This psychophysical task has been used to reliably assess visual information processing (Brown and Crewther, 2017; Ebaid et al., 2017; Ebaid and Crewther, 2019).

Rapid Automatic Naming (RAN) as a Measure of Visual Object Verbalization

The RAN of familiar objects task is a task requiring visual object verbalization. RAN has long been considered a predictor of reading ability (Denckla and Rudel, 1974; Georgiou et al., 2013; Siddaiah and Padakannaya, 2015). RAN has been regularly used

in reading research since first introduced by Denckla and Rudel (1974) and is widely used in children aged 4–10 with test-retest reliability $r = 0.77$ (Denckla and Rudel, 1974; Wagner et al., 1999; Crewther et al., 2011, 2017; Savage et al., 2018; Barutchu et al., 2020; Peters et al., 2020). RAN of objects was chosen as a measure of how fast and accurately a participant could verbally name all 36 everyday objects shown on one A4 sheet. The task began with a practice trial using all objects (boat, star, pencil, chair, fish, and key), to ensure that each participant was familiar with all objects and the agreed name. The participants were then instructed to sequentially name, as quickly as possible, the series of nine objects in each of the four rows starting in the top left corner. The time taken to name all objects was recorded using a stopwatch (see **Figure 2**).

Slurp (Rate of Visuomotor Processing)

The Lee-Ryan Eye-Hand Coordination Test (SLURP) was used to test the development of the visuomotor rate of information processing. SLURP is an iPad application developed by Lee et al. (2014) and designed by Malcolm Ryan to assess eye-hand coordination in terms of accuracy (number of errors) and time. This novel task has been demonstrated to be reliable and valid data and is normed for populations (5–88 years) across the lifespan (Junghans and Khuu, 2019). The task is game-like and requires children to trace shapes with their fingers as quickly and accurately as possible. The task begins with the Castle shape as a practice trial and then five shapes for the actual task in the following order (Circle, Tringle, Square, Rabbit and Snail). Slurp is a task that requires a motor response that involves vision and sustained visual attention to accomplish the task (see **Figure 3**). The total task duration is approximately 2 min.

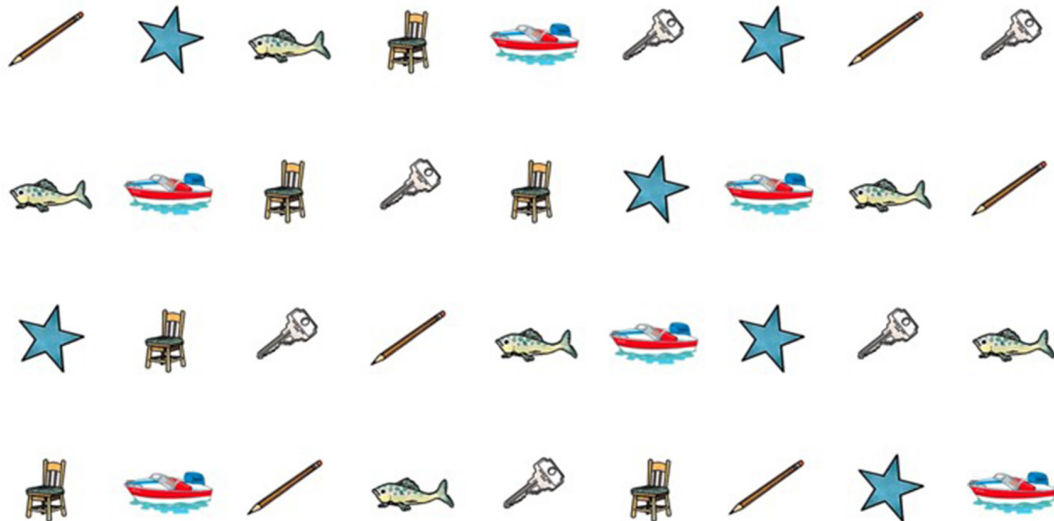
A**B**

FIGURE 2 | Visual object verbalization, rapid automatized naming (RAN). **(A)** Practice trial, **(B)** timed task.



FIGURE 3 | Example of Rabbit shape requiring visually guided tracing in the SLURP task.

Procedure

All testing sessions were conducted during school hours in a quiet room on school grounds. Each session was limited to 30 min maximum and varied according to the child's attention span, interest, and motivation. Participants were asked which game they preferred to start with; the iPad (SLURP) or the computer game (IT). Short breaks were provided when requested and in between tasks. Reinforcements in the form of praise were provided to children at the end of each task.

Data Analysis

All data were analyzed using IBM SPSS statistics version 26. The data were checked for normality according to Shapiro-Wilk's test ($p > 0.05$) with two outliers being identified and removed following inspection of boxplots. Pearson product-moment correlation analysis was used to explore the relationships between nonverbal IQ, chronological age CA, months at school (MAS) and the three experimental measures of rates of visual processing. For initial analysis assessing developmental changes in nonverbal

IQ and cognitive processing task performance, participants were divided into three chronological age (CA) groups (5, 6 and 7 years; see demographics **Table 1**) and then into school Grades assuming this would differentiate children by 1 year of formal schooling (Prep, Grade 1 and Grade 2). Following assumption testing, one-way ANOVAs were conducted to determine whether performance differed significantly on the nonverbal IQ (RCPM), visual, visual-verbal processing tasks (IT and RAN) and the SLURP visuomotor across age groups and grade levels.

Hierarchical regressions were then conducted to investigate the degree to which the length of time attending formal school and presumably studying the domain-specific measures of visual-verbal information processing that is associated with reading and visuomotor (eye-hand co-ordination) processing as necessary for writing affects nonverbal IQ in children aged between 5 and 7 years. Measures of visual information processing were entered at step one to investigate each task's specific contribution to predicting performance on the nonverbal IQ. Age and MAS were entered at step two to investigate the additional contribution to nonverbal IQ beyond visual, visual-verbal processing and visuomotor performance. Path analysis was then performed in order to further examine the hypothesis regarding the mediating effect of age-related development of visual processing speed on nonverbal IQ using the PROCESS SPSS Macro (Hayes, 2017). All analyses were conducted with an alpha of $p < 0.05$ level of significance.

RESULTS

Relationships Between Age and School Years, With Nonverbal IQ, Visual, Visual-Verbal Processing, and Visuomotor Performance

Pearson correlations presented in **Table 2** demonstrated that Age (years) and MAS were significantly negatively correlated with RAN and SLURP task duration, indicating that time required to complete the tasks decreased with age and months of schooling. The relationships between nonverbal IQ raw scores and total time to complete the visual-verbal and visual-motor task were both moderate and negative as accurate performance became faster in both visually driven RAN for objects (reading-like) and SLURP (writing-like) activities with less time being required for completion especially in terms of months at school. The relationships between SLURP and RAN were moderate and positive ($r = 0.411$).

Differences in Nonverbal IQ and Rate of Processing Task Performance Across Age and Grades

To determine the age-related changes and the grades differences in nonverbal IQ (RCPM), visual processing (as measured by IT), visual-verbal processing (RAN), and visuomotor performance (SLURP) across the three age groups, 5 years, 6 years, and 7 years, a series of one-way ANOVAs were conducted (**Figure 4**). Group sizes, age and descriptive statistics for all dependent measures are shown in **Table 3**.

The second ANOVA showed significant differences between the three grades for nonverbal IQ ($F_{(2,98)} = 29.531$, $p < 0.000$), RAN ($F_{(2,53)} = 6.736$, $p < 0.002$) and SLURP ($F_{(2,96)} = 13.351$, $p < 0.000$), as scores in these measures improved significantly with advancing grade level. *Post hoc* tests revealed that the Grade 1 and 2 students had significantly higher raw IQ scores and better performance on the Slurp task compared to the Prep. Further, the Grade 2 performed significantly in the RAN task compared to the Prep students (see **Figure 5**).

Contribution of Visual Processing Speed, Age and MAS in Accounting for Nonverbal IQ

A series of multiple hierarchical regressions were conducted to determine how much each of the three visual domains (visual, visual-verbal and visuomotor) being investigated contribute to nonverbal IQ, and whether age or MAS contribute more to performance on the nonverbal IQ after controlling for task performance. The assumptions of multicollinearity, linearity, and homoscedasticity were not violated. **Table 4** depicts the hierarchical regression outcomes for measures of speed of visual processing, age and MAS in predicting nonverbal IQ, respectively.

Regression analyses reveal that at the ages under investigation, visual processing measures and age significantly predict nonverbal IQ. The total contribution of visual processing tasks (IT, RAN, and SLURP) and age to predicting nonverbal IQ was 43%, with visual processing tasks accounting for 25% of the variance and age adding 18% of the variation. Examination of individual predictors revealed that RAN (visual-verbal processing) was the only significant predictor of nonverbal IQ in the first step, accounting for 8% of the variation whereas age in the second step accounted for 18%. Although chronological age was a significant predictor of nonverbal intelligence beyond visual processing measures, the second analysis demonstrates that MAS provides a further contribution to nonverbal IQ. Analysis of the contribution of visual processing speed tasks and MAS to nonverbal IQ was significant. The addition of MAS significantly accounted for a further 21% of variance beyond the contribution of visual processing measures (27% of variance), with both levels of the hierarchical regressions explaining 48% of the total variance in nonverbal IQ. The unique contribution of MAS to nonverbal IQ was larger than age, 20% and 18% respectively.

To further determine the mediating effect of age on processing speed and nonverbal IQ a path analysis was conducted (**Figure 6**). The regressions paths for both visual processing speed measures (IT and RAN) on age were significant ($b = -0.25$, $se = 0.002$, $p = 0.017$ and $b = -0.41$, $se = 0.008$, $p = 0.001$), respectively. However, the regression of visuomotor performance assessed with SLURP on age was not significant ($b = -0.14$; $se = 0.005$; $p = 0.197$). The regression from the mediator (age) to nonverbal IQ was significant ($b = -0.65$; $se = 0.509$; $p = 0.001$). Based on 10,000 bootstrap samples (MacKinnon et al., 2004) a bias-corrected bootstrap confidence interval for the indirect effect of nonverbal IQ did not contain zero through age. The

TABLE 2 | Correlation (Pearson's *r*) between age, months at school, nonverbal intelligence, visual processing, visual-verbal processing, and visuomotor skills.

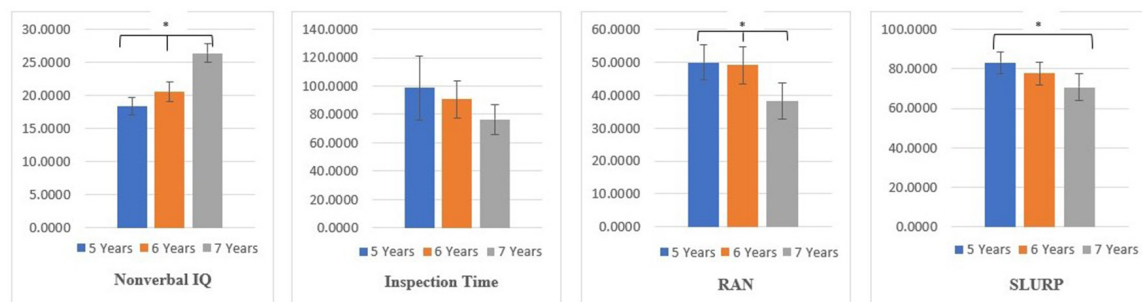
Measure	2.	3.	4.	5.	6.
1. Age	0.905**	0.650**	−0.163	−0.427**	−0.348**
2. Months at school	–	0.618**	−0.055	−0.510**	−0.385**
3. IQ	–	–	−0.102	−0.315*	−0.304**
4. IT	–	–	–	0.157	−0.195
5. RAN	–	–	–	–	0.411**
6. SLURP	–	–	–	–	–

Note. ** $p \leq 0.01$, * $p \leq 0.05$. IQ raw scores, Raven's Coloured Progressive Matrices task; IT, Inspection Time; RAN, Rapid Automatized Naming visual object verbalization; SLURP, total duration in seconds.

TABLE 3 | Descriptive statistics for visual, visual/verbal, and visuomotor processing by age and grade.

		M	SD		M	SD
Nonverbal IQ (raw scores)	5 years	18.37	3.64	Prep	19.18	4.08
	6 years	20.54	4.54	Grade 1	24.89	4.53
	7 years	26.37	3.88	Grade 2	26.50	3.98
Inspection Time (ms)	5 years	98	60	Prep	91	51
	6 years	90	40	Grade 1	86	34
	7 years	76	30	Grade 2	70	14
RAN (Seconds)	5 years	49.96	12.03	Prep	51.61	12.27
	6 years	49.12	12.51	Grade 1	42.34	9.73
	7 years	38.19	9.55	Grade 2	38.19	8.74
Total duration SLURP (Seconds)	5 years	83.17	15.10	Prep	81.47	15.53
	6 years	77.73	17.27	Grade 1	70.36	15.66
	7 years	70.70	18.36	Grade 2	64.10	12.90

ANOVA revealed significant differences between age groups for nonverbal IQ ($F_{(2,96)} = 31.061$, $p < 0.000$), RAN ($F_{(2,49)} = 4.353$, $p < 0.018$), and SLURP tasks ($F_{(2,94)} = 4.071$, $p < 0.020$), as scores in these measures increased significantly over time in years. Post hoc tests revealed that the 7-year-old group had significantly higher raw IQ scores and faster performance on the RAN task compared to the 5- and 6-year old age groups. Furthermore, the 7-year old group was significantly faster at completing the SLURP task compared to the 5-year old group. While the rate of visual processing appeared to decrease across age groups in the IT task, this change was not significant $F_{(2,90)} = 1.946$, $p \leq 0.149$ (see Figure 4).

**FIGURE 4 |** Age group differences (+/- 95% Confidence Intervals) for, nonverbal IQ, rate of visual information processing (Inspection Time), visual-verbal processing (RNA), and visuomotor processing (SLURP). Note. * $p < 0.05$.

indirect effect of visual processing speed measures IT, RAN, and SLURP to nonverbal IQ through age was not significant (−0.16, −0.27, and −0.10), respectively.

DISCUSSION

This study aimed to investigate the contribution of age-related and school experience related differences in speed of information processing of visual, visual-verbal, visuomotor dual tasks and their relationships to nonverbal IQ of children aged 5–7 years old. We also aimed to examine the contribution of age and domain-specific school influences on the rate of processing

to nonverbal IQ. The key findings were that nonverbal IQ was significantly correlated with age and MAS, and negatively correlated with the rate of sensory information processing (visual-verbal and visuomotor). Performance of the 7-year-old group showed a significant increase beyond that of the 5- and 6-years groups on measures of visual-verbal information processing assessed by RAN task, visuomotor skills tested by SLURP and nonverbal IQ, but not on simple visual information processing (IT task), for which there was a decreasing nonsignificant trend in threshold exposure time needed for accurate object identification. Grade 1 and 2 who attended formal schooling for 1 and 2 years longer than the

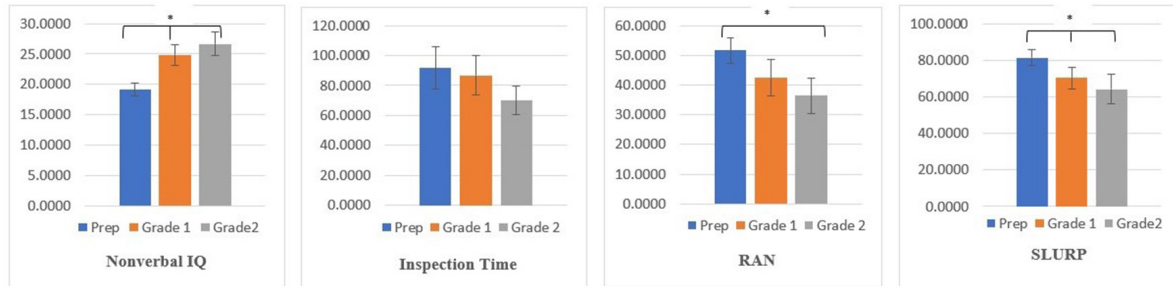


FIGURE 5 | Grades means differences (+/- 95% Confidence Intervals) for, nonverbal IQ, rate of visual information processing (Inspection Time), visual-verbal processing (RNA), and visuomotor processing (SLURP). Note. * $p \leq 0.05$.

Prep students, respectively, performed significantly better on RAN and SLURP tasks. Finally, MAS was associated with greater contributions to nonverbal intelligence than chronological age which supports the increases in domain-specific rate of processing.

Relationships Between Nonverbal Intelligence and Sensory Information Processing

Consistent with our hypotheses and past research, nonverbal reasoning was significantly negatively correlated to the speed of visual-verbal and visuomotor information processing but not to simple visual perceptual task speed (Nettelbeck and Young, 1990; Coyle et al., 2011; Demetriou et al., 2014) with decreases in processing time being associated with increases

in performance on nonverbal intelligence tests. Nettelbeck and Young (1990) examined the relationship between fluid intelligence (assessed on the Weschler Intelligence Scale for Children) and visual non-motor information processing speed in 6–7-year olds and found a moderate negative correlation (-0.31) similar to the findings of the current study. Furthermore, Kail (1991a) and Kail and Hall (1994) have also long observed that processing speed with motor components contributes significantly to intelligence. Indeed, Kail (2000) postulated that processing speed *per se* may be a great predictor of intelligence even in infancy. The efficiency of given information processing has also been noted to play a critical role in individual developmental differences of general intelligence (Demetriou et al., 2013). In terms of the visual perceptual and nonverbal IQ, the results show nonsignificant relationships between our IT task and nonverbal intelligence though a

TABLE 4 | Predictive contribution of visual, visual-verbal processing, and visuomotor skills, age and MAS on nonverbal IQ.

Variable		Nonverbal IQ		
		β	R	sr
Step 1				
	IT	−0.22	0.50	−0.21
	RNA	−0.34*	0.50	−0.29
	SLURP	−0.21	0.50	−0.18
$R^2 = 0.20$; F change (3,44) = 4.848; $p = 0.005$				
Step 2				
	IT	−0.10	0.50	−0.098
	RAN	−0.11	0.50	−0.87
	SLURP	−0.11	0.50	−0.095
	Age	0.51*	0.65	0.424
$R^2 = 0.38$, Change $R^2 = 0.18$; F change (1,43) = 13.516; $p = 0.001$ Total $R^2 = 0.43$; F (4,43) = 8.049				
Step 1				
	IT	−0.26	0.52	−0.18
	RAN	−0.39	0.52	−0.32
	SLURP	−0.17	0.52	−0.14
$R^2 = 0.22$; F change (3,43) = 5.377; $p = 0.003$				
Step 2				
	IT	−0.19	0.52	−0.18
	RAN	−0.08	0.52	−0.061
	SLURP	−0.11	0.52	−0.090
		0.56*	0.69	0.454
$R^2 = 0.43$, Change $R^2 = 0.21$; F change (1,42) = 16.605; $p = 0.000$ Total $R^2 = 0.48$; F (4,42) = 9.648				

Note. IT, Inspection Time; RAN, Rapid Automatic Naming; SLURP, visuomotor performance task; Nonverbal IQ, Intelligence Quotient score on Raven's Coloured Progressive Matrices task; MAS, Months at School. * $p \geq 0.01$; according to Cohen's guidelines, $r \geq 0.10$, $r \geq 0.30$, and $r \geq 0.50$, represent small, medium, and large effect sizes, respectively (Cohen, 2013).

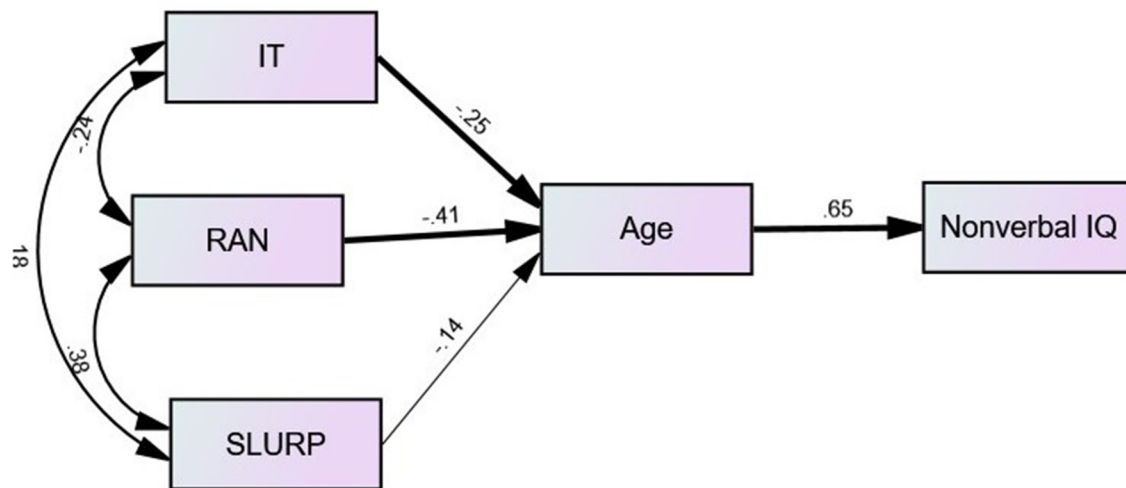


FIGURE 6 | Path-diagram of age mediating the relationships between visual processing speed and nonverbal IQ. IT, Inspection Time; RAN, Rapid Automatic Naming; SLURP, visuomotor performance task; Nonverbal IQ, Intelligence Quotient score on Raven's Coloured Progressive Matrices task. *Note.* Significant paths are in bold.

gradually increasing trend in performance of both which is in line with previous research investigating their relationship and suggesting insignificant and weak correlations especially in school age children (Smith and Stanley, 1983; Anderson, 1988, 1989; Mackenzie et al., 1991; Dandy, 2000). Many of these foregoing studies were included in a meta-analysis conducted by Grudnik and Kranzler (2001) and led to the conclusion that the association between IT tasks and IQ is comparable within the included studies. Jensen (2006) suggested a constant relationship between IT tasks and measures of IQ, which stands in contrast with our results even for a rank-order correlation between IT and nonverbal IQ that could be due to large variability in performance for our youngest children. Together, all these earlier studies in combination with the results presented here support the hypothesis that motor speed of information processing and fluid intelligence develop concurrently with age, and the corollary that fluid intelligence is usually accepted as a function of the rate of information processing.

Visual, Visual-Verbal Information Processing and Its Relationship With Visuomotor Performance

Overall, our findings demonstrate a moderate and positive relationship ($r = 0.356$) between visual/verbal processing and visuomotor performance in children aged 5–7 years old while controlling for chronological age. Son and Meisels (2006) have previously reported a moderate positive relationship between cognitive skills and visual-motor skills in a longitudinal study of kindergarteners and Grade 1, $r = 0.35$ and $r = 0.40$, respectively. Further studies concur with our results, which present moderate to large associations between total score of cognitive abilities and fine motor performance in children aged 6–8 years old (Abdelkarim et al., 2017), and 4–11 years old (Davis et al.,

2011). Our findings also lend support to previous literature indicating that cognitive and motor skills develop along the same timeline in children aged 5–10 years, though the strength of the relationship is less stable beyond 10 years of age (Anderson et al., 2001). Collectively, these findings indicate that children who perform well on cognitive skill tasks (fluid intelligence, visual processing, response inhibition, attention and working memory), are also more likely to perform well on visuomotor tasks (van der Fels et al., 2015). This significant association between both dual visually-driven motor measured functions (cognitive and fine motor) could be explained neuroanatomically as they share similar Magnocellular-driven attention and dorsal brain networks (Crewther et al., 1999; Laycock et al., 2007) and regions of interest (Leisman et al., 2016). Indeed the maturation of the dorsal visual stream (Goodale and Milner, 1992) and the dorsal streams' dorsal and ventral goal-directed parieto-frontal pathways are thought to be responsible for visually driven attention action and goal directed behaviors and working memory (Corbetta and Shulman, 2002; Rizzo et al., 2017).

Simple visual information processing speed assessed with the IT task tended to increase with age but not at a rate that significantly correlated with visuomotor performance assessed by SLURP. This is in line with previous results by Ebaid et al. (2017) who investigated the relationship between visuomotor integration using Pegboard and processing speed measures (IT) in young and older adults and also found no significant relationships.

Age/Grades and Performance on Nonverbal IQ and Measures of Visual Information Processing

As hypothesized, nonverbal IQ raw scores significantly increased with age and years of schooling, and the rate of visual-verbal

processing assessed by RAN and visuomotor performance tested by SLURP decreased with age and length of time attending school. Our findings indicate that visually assessed nonverbal IQ and visual sensory information processing (visual-verbal and visual-motor) develop concurrently as children mature and become faster in processing and responding to sensory information, which in turn improves their reasoning capacities as evidenced by performance on tests of nonverbal intelligence such as RCPM (Fry and Hale, 2000; Kail, 2000; Cotton et al., 2005).

Visual-verbal processing and visuomotor performance are anecdotally reported to change significantly in children 5–7 years old when they begin formal schooling and start formally practising and utilizing their reading out loud and writing skills. Our findings also demonstrate that threshold IT continues to trend downward with age, though not to the point of reaching a significant level among the three age groups (5, 6, and 7-year-olds). This may be attributable to structural immaturity of the visual pathway projections (Hendrickson and Drucker, 1992; Crewther et al., 1999) that are not fully developed until late childhood/early adolescent years (Crewther et al., 1999; Hendrickson et al., 2012), in accordance with the functional development of the magnocellular (M) fast and parvocellular (P) slower projections to the visual cortex (Klistorner et al., 1997; Leat et al., 2009). Again, this is most likely because IT would be expected to be related to morphological maturation of the fovea of the retina around 5–6 years of age (Hendrickson and Drucker, 1992). By comparison, speed of processing of more complex dual visual tasks that are partially dependent on verbal or manual motor reaction time would be expected to initiate higher cognitive demands (selection and inhibition) and be highly affected by chronological age early in life (Nettelbeck and Wilson, 1985; Anderson, 1989; Nettelbeck and Young, 1989; Anderson et al., 2001).

Findings from the current study are consistent with previous results that have utilized speeded dual motor component tasks (time measurements) namely; perceptual motor tasks, the Tapping task, the Pegboard task, (Kail, 1991b) naming speed tasks, the coding task from WISC (Kail and Hall, 1994), RAN (Neuhaus et al., 2001), and response time tasks (Miller and Vernon, 1997) in children and confirm significant age differences in these tasks among children aged 4–8 years (Cotton et al., 2005). Our results share a number of similarities with these studies indicating that response time decreases as age increases and that the rate of change in information processing is faster in childhood.

Contributions of Months of Early Schooling to Rates of Visual Information Processing and Nonverbal IQ

Our analyses demonstrate a positive contribution of schooling to the rate of visual information processing namely visual-verbal and visuomotor performance which is a reflection of the experience and practice of reading and writing once the child enters a formalized schooling system. This result is

in line with Alexander and Martin (2004) who investigated the effect of schooling on cognitive abilities and suggested a greater influence of schooling than chronological age on verbal processing tasks, that are associated with reading ability. fMRI studies investigating the role of schooling 5–7 years children on brain function have demonstrated that practise and experience play a key role in brain activation, especially in the right posterior parietal cortex, that is associated with control of eye movements and shifts in attention (Wurtz and Goldberg, 1972), and executive function improvement (Burrage et al., 2008; Brod et al., 2017; Morrison et al., 2019). Similarly, Morrison et al. (2019), who reviewed the “casual” impact of schooling on cognition in school beginners (Pre-kindergarten, kindergarten, and Grade 1) demonstrated a strong impact on a variety of cognitive processing skills (attention control and working memory) that are essential for successful reading (visual-verbal) and writing (visuomotor integration). Our results are also in line with Duan et al. (2010) which highlight the more important role of knowledge and experience than age maturation in the speed of information processing development in children aged 9–13. Although age and time attending formal schooling are confounded, some studies have solved this issue by comparing two groups of students (Prep and Grade1) at the same chronological age, but where one group enrolled at school earlier than the other, and have also shown that 1 year of schooling has a stronger influence on cognitive functions namely, processing speed, sustained attention, working memory, cognitive flexibility, spatial ability, and inhibitory control than chronological age alone (Dasen et al., 2004; Burrage et al., 2008; Brod et al., 2017).

Our regression analyses supported the influence of domain-specific knowledge on visual processing development that is associated with practising of reading and writing abilities at schools. This related increase in the rate of visual-verbal (reading) and visuomotor performance (writing) due to formalized schooling significantly contributes to nonverbal IQ. Our results are in agreement with a wide range of studies that have compared the contribution of age and months of schooling effect to intellectual ability and suggested a greater contribution of schooling than age (Ceci, 1991; Artman et al., 2006; Cliffordson and Gustafsson, 2008; Brinch and Galloway, 2012; Ritchie et al., 2013). According to Cliffordson and Gustafsson (2008), months attending formal schooling significantly contributed to children’s performance on general intelligence tasks not only specific knowledge abilities that improve with regular formalized practice at school. Lastly, these studies have highlighted the usefulness of RAN as an early correlate measure of nonverbal IQ, and as a well-established predictive measure of potential reading ability in preschool and early readers (Anthony et al., 2007; Furnes and Samuelsson, 2009; Fricke et al., 2016; Peters et al., 2020).

CONCLUSIONS

Overall, this study has demonstrated that the acquisition of more complex visually based skills such as visual-verbal and visual-motor information processing and nonverbal IQ, develop

concurrently during the early school years. Visual-verbal and visuomotor processing correlated, significantly though simple visual processing assessed with thresholds IT task did not reach significance level with any variables. Regression analyses comparing the prediction of age vs. MAS beyond domain-specific rates of processing to nonverbal IQ indicate a larger contribution of MAS than chronological age to nonverbal IQ. Hence, our results support the domain-specific hypothesis demonstrating that months attending formal school contribute significantly more to cognitive performance than age, i.e., reading and writing associated abilities improve rapidly with regular practice (Burrage et al., 2008; Brod et al., 2017; Morrison et al., 2019), rather than the rate of simple visual information processing *per se* that is well developed even when starting school (Klistorner et al., 1997; Leat et al., 2009). Most importantly, the findings of this study provide further evidence that measures of rates of information processing in RAN and the SLURP are suitable measures for early identification of children likely to score lower in nonverbal IQ tests and have difficulties learning to read. However, further work with a larger sample size needs to be performed to determine the developmental changes of the rate of sensory information processing across wider age groups.

DATA AVAILABILITY STATEMENT

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

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ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the La Trobe University Human Ethics Committee, the Victorian Department of Education Human Ethics Committee, and the Victorian Catholic Schools Ethics Committee. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

RA and SC designed this study, and interpreted the data with MM. Both RA and MM conducted the data analysis and all authors contributed to writing up of the study. RA was assisted in data collection by Ms. Hayley Pickering and Ms. Areej Alhamdan. All authors contributed to the article and approved the submitted version.

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The First Year Developmental Crisis: Origin of Cultural Action

Yulia Solovieva^{1,2*} and Luis Quintanar²

¹ Meritorious Autonomous University of Puebla, Puebla, Mexico, ² Autonomous University of Tlaxcala, Tlaxcala, Mexico

According to the conception of cultural historical psychology, introduced by L. S. Vigotsky, the first year of a child's life represents a specific period of development or the first psychological age. Psychological development should be differentiated from biological development and have proper objective indicators. Psychological development starts with the possibility of initial cultural communication between an adult and child, within a unique kind of social situation of development. The goal of the article is to describe the content of the crisis of the first year of life as a psychological phenomenon and to propose psychological and neuropsychological indicators for qualitative assessment of the progress of psychological development at the end of the first year of life. The article opens the discussion about guiding the activity of the first age, new psychological formations of the crisis of the first year, and qualitative changes in the social situation of development. The content of the first psychological age and crisis is presented in the article, according to proposals of cultural historical psychology. Future interdisciplinary research should be continued in order to establish optimal strategies for adult and child interaction during a stable period of development and the crisis of the first year.

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*Correspondence:

Yulia Solovieva
yulia.solovieva@correo.buap.mx

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"Relation between the child and reality, from the very beginning, is social relation. In his sense, the human newborn is the social being by excellency [sic]."

L. S. Vigotsky

INTRODUCTION

The first year of life should be understood as a specific period of development or as a particular psychological age, including a stable period of development with specific phases and the critical period. This period has its own content and structure, quite different from the content of later psychological ages. Psychological age cannot be determined only by chronological limits, but has to be analyzed according to developmental manifestations of each child with normal and abnormal development (Vigotsky, 1984).

According to the conception of cultural development, introduced by L. S. Vigotsky in psychology, psychological development cannot be reduced to biological development, physiological processes, or neurological maturation (Leontiev, 2009; Lisina, 2009). Psychological age is a period of ontogenetic development characterized by proper manifestation of a child's activity and personality. Central and accessory lines of development exist within the guiding activity of the age. Psychological age includes specific guiding activity, which leads to gradual acquisition of new psychological formations (Elkonin, 1995). New psychological formations might be acquired as a result of constant joint activity between adult and child. Such joint activity occurs in conditions of specific social situations of development. New psychological formations appear at a critical period and reflect

a developmental crisis. New psychological formations are qualitative characteristics of the child's activity and personality, which appear at the end of the age and announce the critical period, as the passage to the next period of age (Vigotsky, 1984; Obukhova, 2006). According to Bozhovich (1981, pp. 130, 131), "new psychological formation appears as a specific sequence, which characterize[s] the periods of central line of their ontogenetic development." We suppose that the study of this sequence of new psychological formation in infancy has not yet been completed.

In each case of psychological development, a social situation of development might be negative or positive in relation to the guiding activity of the child. Social situations of development might be understood as the actions of the adult directed to the child during each period of development. These actions might be favorable or unfavorable for guiding the activity of psychological age (Solovieva and Quintanar, 2020).

Vigotsky (1984) has written that the problem of psychological age is the central problem of psychological research and of psychological assessment. The task of developmental psychology is to study the content of each psychological age in positive and negative social and organic conditions of development.

Psychological development starts with the possibility of initial cultural communication between an adult and child, within a unique kind of social situation of development. According to cultural historical perspective in psychology, an adult is a representative of cultural experience, which has to pass from them to the child (Obukhova, 2006; Ilienkov, 2009). The child's

activity is not an individual activity from the very beginning, but is an act of communicative collaboration with an adult. The adult's activity, directed to the child, is the unique origin of a child's cultural development.

Each psychological age might be studied and assessed according to the terms of social situations of development, guiding activity, new psychological formations, central and accessory lines of development, and the crisis of the age as the bridge for the next period of development. **Table 1** shows the structure of each psychological age.

The concrete content of psychological and neuropsychological assessment of stable and critical periods of development is one of the essential problems in developmental psychology in cases of children with optimal development and with difficulties (Solovieva and Quintanar, 2016).

The goal of the article is to describe the content of the crisis of the first year of life as a psychological phenomenon and to propose psychological and neuropsychological indicators for qualitative assessment of progress of psychological development at the end of the first year of life. The main concepts introduced by Vigotsky (1984) and his followers (Leontiev, 1984, 2003; Elkonin, 1980, 1995; Lisina, 2009) are used as theoretical background for precision of our ideas.

THE CONTENT OF THE FIRST PSYCHOLOGICAL AGE

Let us revise the structure of the first psychological age. Guiding activity during this period might be understood as activity of communication between adult and child. Communication is not a natural expression of a child's individual life, but a kind of cultural activity that has its own motives, goals, means, results, and operations (Lisina, 2009). This kind of activity has to be gradually formed from an external level of social communication into an internal level of the child's activity. Bozhovich (1981) stresses that the consciousness of the baby at the beginning of the first year of life represents mostly emotional components, related to immediate interactions.

How does this communication start, as it is obvious that the child is born without any kind of cultural communication and even language would appear as the result of development at the end of the first year of life, in cases of possible development? How is it possible to convert some kinds of diffuse immediate interactions, related to the emotions of the child, into meaningful process of one's own cultural activity?

Cultural communication starts with the clear expression of communicative intention. Commonly, it is possible to notice this intention between the end of the first month and the third month of a child's life. From the age of five months, such complex communication, with notable expression of a child's smile directed to an adult's face, might be consolidated (Bazhenova et al., 2007).

Initial intention for communication, represented as a complex of animation, includes four essential elements, which might be identified in the situation of communication between adult and child. We propose to assess these elements as follows:

TABLE 1 | The structure of psychological age.

Element of the structure of psychological age	Description	Indicators/agents
Social situation of development	Type of relations between the child and society in each age	Actions and attitudes of adults and institution in relation to the child
Guiding activity of the age	Psychological activity, which leads to development in this age	Motivated activity of the child; interests and intentions of the child
Central line of development	Achievements of development during the age	Affective or practical experience of the child as new manifestations of development
Accessory line of development	Achievement of previous age or conformation of the basis for the next age	Affective or practical experience of the child as basis of development
New psychological formations	New qualitative features of activity and personality at the end of the age	Concrete indicators of development, which might be discovered by qualitative assessment
Stable phases	Long period of development within the same social situation	Observation of social situation of development and guiding activity
Critical phase	Brief manifestation of the need for changing of social situation of development	Psychological assessment of new formations of the age with observation of social situation of development and guiding activity



FIGURE 1 | Complex of animation during the first months of life.¹

- 1 Exchanging of mutual eye contact between adult and child.
- 2 General non-specific corporal agitation and movements of arms and legs of the child in direction to the adult.
- 3 Smile of the child in response to the smile of the adult.
- 4 Non-specific vocal expression of the child in response to the actions and speech of the adult.

All these four elements should appear simultaneously and in the presence of the adult, together with the whole situation of joint communication with the adult. Isolated movements, such as sounds or even a smile as reaction to physiological satisfaction, are not indicators of animation complex. The presence of the mentioned components of animation complex might be identified only in the situation of adult and child communication, so that psychological assessment in this age should be conducted only in the presence of the close adult in the situation of close affective communication. **Figure 1** presents a clear example of such a situation.

All these elements conform to the famous animation complex, introduced in literature by Lisina (1986, 2009). The animation complex can be defined as an indicator of the start of a child's communication activity with proper intention; this intention becomes conscious at the end of the first year of life. The periods of duration of animation complex may be several minutes long and is expressed mainly between the second and eight months of a child's life. After that, the animation complex disappears which opens the path for changing of means of communication between adult and child and is characterized by the inclusion of objects and the first manifestation of words or of "autonomous speech" in terms of Vigotsky (1982).

The concept of animation complex is essential for identification of the start of communicative activity as the child's own activity (Lisina, 2009). With communicative expression, the child converts into the subject of proper activity. Vigotsky has written that the newborn child is the most social being and that the whole process of development should be understood as the process of "individualization" of the child's psyche. Such

a position is contrary to the traditional way of understanding development as the process of "socialization" (Piaget, 1976). Different lines of psychological development were proposed by prominent psychologists of the last century: "individualization" (Vigotsky) and "socialization" (Piaget, 1976). It is possible to refer to these positions as two general paradigms: a paradigm of cultural historical development, based on proper activity of psychological subject, and a paradigm of constructivism, based on biological maturation as the feature of natural evolution (Solovieva and Quintanar, 2019).

Communicative activity is the only kind of activity that provides positive appropriation of general cultural experience during the first year. Absence of this activity may have negative consequences in psychological development, such as the absence of appearance of language at the critical period, absence of actions with objects, and absence of independent movements directed to goals.

It is very important to stress that communication would never start automatically from the child, but is a result of an adult's efforts to provide social joint communication. According to Vigotsky (1984), psychological functions of the child are initially divided between an adult and a child. That is why the child's functions are social from the very beginning and become individual only later.

It is important to remember that there is no communicative activity of the child without communicative activity with an adult. At the same time, this activity should be positive affective communication. The indifference of an adult evokes the indifference of the child and their negative attitude provokes a negative response from the child.

Table 2 shows the content of the first psychological age according to general structure of the age.

COMMUNICATIVE ACTIVITY AND ITS BRAIN ORGANIZATION IN THE FIRST YEAR OF LIFE

The adult organizes the life of the child; the adult decides when to move, to carry, to feed, to bathe, to go to sleep, when to go for a walk, or when to play. The adult establishes the goals of the child's communication, provides different objects, and offers the verbal determination of objects and situations. The conformation of functional relations between unions of nervous mechanisms might be organized only as a result of cultural activity directed to previously established goals. The human brain is not the source of psychological functions, but functional level of realization of human cultural activity, which conforms to functional systems (Anokhin, 1980) or functional organs during a child's active life (Leontiev, 2000). Functional systems in humans are not only the result of natural maturation, but also of cultural activities (Bernstein, 2003; Machinskaya, 2012), which emerge as joint activities guided by an adult.

Parameters of the structure of psychological age may serve as a proposal of qualitative assessment of psychological development of the child. The content of the guiding activity might be used for study and assessment of brain mechanisms, which take part in this activity.

¹ Photograph from the authors' family archive.

Communication as a kind of human activity cannot be based on only one isolated “area” of development or only one brain zone or one neural net (Gazzaniga, 1993; Bassett and Gazzaniga, 2011; Dehaene, 2015). Each cultural activity requires functional organization and participation of the whole central nervous system, including three functional units: unit of general brain activation, unit of processing of sensorial external and internal information, and unit of programming and control (Luria, 1969). Developmental neuropsychology intends to discover and follow the formation of such “factors” through diverse periods of ontogenetic development (Solovieva and Quintanar, 2016). The first period of ontogenetic development is the age of joint emotional affective communication between adult and child (Solovieva et al., 2016).

The structure of joint communicative activity includes the social need expressed in concrete motives: an adult, who organized a child's communication. The means or operations of this communication are corporal movements, eye movements, vocalization, and facial expressions of the child. All these means require participation of different levels of hierarchic brain organization, including cortical, subcortical, and cortico-subcortical relations. In this sense, it is not useful to speak about isolated areas of motor development, personality development, and speech development, as communication involves movements, expressions, and vocalizations. The absence of communication results in severe stagnation in a child's

psychological development not only during the first year, but also during further periods of development.

Communicative activity between the adult and child starts with the animation complex. The appearance of this complex can serve as an indicator of appearance of a new functional system, which includes diverse mechanisms of systemic and hierarchic brain organization, or three functional units, according to Luria's (1973) theory.

Firstly, the animation complex requires general non-specific activation of the cortex; general activation includes necessary mechanisms of emotional activation as affective attraction and involvement of the child into the process of communication. All subcortical structures of the brainstem and limbic circuits take part in this general activation (Luria, 1973).

Secondly, the animation complex includes processes of perception of all modalities: auditive (voice of adult), visual (face of adult), and tactile (gentle touching of an adult). Such perception guarantees formation of complex polymodal images and retention of significant information. Posterior zones of the brain cortex take part in this perception.

Thirdly, the whole emotional complex of animation is directed to an adult, who is the motive of this communication activity. The movements of the child's eye and contact with the adult's gaze is the central mechanism of this complex. In this case, the unit of programming and regulation of movements takes part, so that the eyes of the child might concentrate on the face of an adult for longer periods. Frontal lobes with zones of regulation of eye movements, but also of all level of programming of movements, guarantee this process. Essential participation of the anterior frontal (orbital) regions guarantees connections with the limbic circuit and thalamic system, which also provides relations of anterior cortex with all posterior brain zones and zones of processing of sensorial modalities (Bezrukikh and Farber, 2000; Bezrukikh et al., 2009).

This example shows that there is no reason to talk about each part of the brain or about each cognitive process in an isolated manner, for example, about motor reactions, perception, and speech as though they were independent isolated functions (Glozman, 2009; Akhutina and Pilayeva, 2012). Luria (1973) has expressed that it is a great mistake to think that each brain unit works separately for some kinds of cognitive functions, but that the general participation of all units guarantees human complex activity.

Actions of affective communication between adult and child provide visual sustained contact directed to a cultural goal. An important indicator of this sustained contact is the child's smile, directed to the adult's smiling face. It was precisely this child's smile that was noticed by Vigotsky (1984) and studied in details by his followers (Lisina, 2009).

Functional brain organization starts together with animation of complex in the first months of the child's life. The absence or late appearance of the animation complex also means the absence or late appearance of functional brain organization.

Communication permits to guarantee social necessity for new experiences and impressions, for example to share contact with others. According to Tomasello (2013), this necessity should be recognized as a basic human necessity. This social necessity is

TABLE 2 | The content of the first psychological age.

Element of the structure of psychological age	Description	Indicators/agents
Social situation of development	All kinds of situations of close emotional contact between adult and child	Concrete actions and attitudes of adults, which guarantee close affective communication in day-to-day life
Guiding activity of the age	Affective emotional communication with an adult	Interest in an adult and aspiration for contact
Central line of development	Affective emotional positive basis for personality	Concrete affective episodes of communication
Accessory line of development	Practical experience of manipulation with objects and toys	Concrete practical episodes with objects
New psychological formations at the end of the age	Locomotion Autonomous speech Actions with objects	Aspiration to stand up and make passes; intentions of articulation of words; intentions for usage of objects independently
Stable phases	The first year of life as the period of emotional communication with two phases	Phase of communication without objects and with objects, starting from the second half of the first year
Critical phase	Manifestation of independency development	Aspiration of separation from adults for more independent movements and actions; appearance of new age formations

the main difference between development of a human child and superior apes, as was shown in different experiments (Tomasello, 2013). Tomasello (2013) proposes to refer to this necessity as empathy. We agree and believe that empathy is a result of early and proper organization of adult child system of communication, starting with the complex of animation.

Four elements of the content of the complex of animation might be clearly related to elements of functional brain organization. **Table 3** proposes a relation between elements of the complex of animation (Lisina, 2009) and functional brain units, according to Luria's (1973) conception.

According to **Table 3**, it is possible to appreciate broad participation of the central nervous system in the animation complex. Very important is that all these brain mechanisms should appear to work simultaneously and not as isolated processes. These brain functional components, while supporting the animation complex, give origin to the unit of stable general brain activation, unit of sensorial perception, and unit of programming and control. The resting of any of these elements might result in disorganization of the whole functional system for positive affective communication with the adult with different possibilities of negative consequences on present and future psychological development.

At the same time, these four elements of the animation complex might serve as indicators of manifestation of directed activity of the child. It is possible to call them both indicators of psychological development and also of the first manifestation of conformation of the functional system of communication action from a neuropsychological point of view. The complex of animation is a positive indicator of first communication action and first functional system with its clear brain mechanisms.

These indicators depend only on actions and attitudes of an adult toward the child; later, the child shows more and more initiative and will direct his or her own attitudes and communicative actions toward other people and objects. Activity of positive affective communication will produce later important changes in the cognitive and affective "image of the world" of the child (Leontiev, 1984).

The zone of proximate development will turn into the zone of actual development for goals and expressions of communication.

At the end of the first year, the child will be conscious of his or her communicative goals. In this case, the child would have conformed communicative actions directed to conscious goals of affective communication: to get, to show, to share, to possess, and so on. Communicative activity will be the basis for the next guiding activity: actions with cultural objects.

TWO PHASES OF DEVELOPMENT OF COMMUNICATIVE ACTIVITY DURING THE FIRST YEAR

Psychological development of the child during the first year suffers transformations; gradually, the "first sensorial generalizations appear, the baby starts to use elements of words and objects" (Bozhovich, 1981, p. 132). The central line of psychological development of affective representations starts to be accompanied by the accessory line of practical usage of the objects. Let us try to understand how it happens.

According to Lisina (2009), a child's communicative activity possesses the following features, typical for this period of development:

- 1 Affective attention and interest of all actions of the adult.
- 2 Emotional response to each contact and action of an adult toward the child.
- 3 Initiative of the child to follow and to involve an adult in communication.
- 4 Sensibility of a child toward positive attitude of an adult, which he/she manifests to other persons, objects, and the child.

Detailed analysis of these characteristics of a child's communicative activity during the first year of life allows to establish two different phases. Gradually, the child starts to pay attention not only to the adult's face, but also to the objects and toys that the adult uses and shows to the child and names in oral speech. The absence and the presence of cultural objects with cultural meaning and the possibility of their usage in clear practical situations makes the difference between the phases of development of communication. According to Lisina (1986), two phases of development of communication between child and adult might be identified.

The first phase of psychological development during early age might be characterized by "communication in personal situations." The first indicator of this activity is a complex of animation, based on exchanging of eye contact and long contact between adult and child, exchanges of smiles together with vocalizations, and general motor excitation of the child toward the appearance and communicative actions of an adult. Such communication is the first guiding activity of cultural development of the child. In cases of positive social situation of development, when an adult is able to guarantee and to start this communication, it starts at the end of the first or second month of a child's life. The positive social situation of development implies constant communicative actions of the adult toward the child. Communication of the adult is always verbal and

TABLE 3 | Animation complex and brain units.

Elements of animation complex	Description	Reference to brain functional organization
Eye's contact	Concentration of the adult's face and eyes	Frontal lobes, area of eye movements; posterior visual primary and secondary zones
Corporal agitation	Movements of arms and legs toward the adult	Motor primary and secondary zones, subcortical structures of organization of movements and muscle tone regulation
Smile	Response to the adult's smile	Frontal lobes and limbic structures
Vocalization	Intents of articulation or emission of guttural sounds	Parietal and auditory sensory and secondary zones of both hemispheres

emotionally expressive, while the communication of the child is only emotionally expressive. All movements of the child are included in the content of this personal affective non-verbal communication with an adult. Within this activity, the adult presents and tries to put different attractive objects and toys in the child's hand. The objects are not interesting for the child *per se*, but only as the objects possessed by an adult.

The cultural object and the possibility of touching and moving the objects opens new qualitative possibilities for the child's development.

The second phase of the first year of life occurs as "practical communication" organized between adult and child. At this period, the child is already able to sit independently and can start to explore and manipulate the objects by himself/herself. The child starts to be fascinated by the appearance of new objects and starts to manipulate them. It even seems that the child is "obsessed" by objects (Lisina, 2009). The child needs constant help and participation of an adult in order to know what to do with different objects. The child also needs the adult to share affective communication and re-affirmation of their own practical intention. Affective communication is still the guiding activity of the age, but new means were already introduced in this activity. These means are cultural objects, offered and used by the adult and directed to the child. Practical initiative and orientation of the adult is an essential condition for further psychological development of each child in human society.

Affective communication in practical situations implies a variety of social situations for collaboration. Each adult should be conscious of the necessity of constant useful propositions of joint actions with the child. Suitable situations for these joint actions might be as follows: going for a walk, election and changing of clothes, admiration of flowers and animals, usage of a spoon, playing with a toy. The time that an adult spends with the child should be directed to the goals of positive affective communication and initiative for stating of practical usage of cultural objects of day-to-day life. It is essential to notice that first manipulations with objects starts before the child may speak and before the child may walk, so that manipulation with cultural objects is the second line of a child's developmental, after the line of shared affective communication. Vigotsky (1982) was right when he wrote that emotions are *alfa* and *omega* of the whole process of development.

Cultural objects are not simply physical objects which have dimensions and sizes. Each cultural object is a product of the history of humanity and of constant production and usage of the objects (Obukhova, 2019). The social situation of development of the first year should consist of an adult's actions, which guarantee both affective and practical involvement of the child into joint realization of communicative and simple practical actions. Alteration of this situation of development results in stagnation on a child's development as absences of activity directed to the goals.

All forms of a child's initiative should be approved by adults. Social initiative and positive approval are the basis for social motives of activity and the basis for the motive of cooperative activity and mutual help (Tomasello et al., 2005). Tomasello (2013) has affirmed that superior apes are not eager to help each

other or another participant of an action, while 9-month-old babies with positive development show motivation for helping and cooperation. According to activity theory, motives of activity emerge as previously established and as shared goals of the same activity (Leontiev, 1984).

In the case of communication activity, the motives for communication might emerge as previously established goals of communication. For example, the adult might direct the attention of the child to an interesting object, such as the moon in the sky. In this case, the moon is a goal of the adult's communication toward the child. Later on, the child would search for the moon in the sky and show it to the adult. In this case, the moon was the motive of communication of the child.

In order to provide meaningful relation of the child with the objects of the cultural world, the adult should introduce meaning to all joint actions. The goals of these actions will turn into conscious intentions and conscious goals of the child's communication.

The content of the consciousness of the child for the end of the first year of life would include their own goals of positive communication. The crisis of the first year of life starts when the child starts to wish, unconsciously, to use and to possess independently more and more new interesting objects. The child even starts to show rejection of an adult's help.

THE CONTENT OF THE CRISIS OF THE FIRST YEAR

The crisis of development at the end of the first year is the result of the whole process of development of communicative activity in its two phases. This crisis serves as an indicator of positive qualitative change in a child's activity and personality. The crisis is expressed by a child's intentions for independent locomotion, articulation of first words, and the performance of actions with cultural objects and toys. This last point is the central new psychological formation of the first age of development. If manifestations of locomotion and articulation are well known in psychology, very little research is dedicated to identification and promotion of development of actions with objects (Solovieva et al., 2018, 2020).

The main feature of the content of the crisis is that the child starts to use the objects according to their cultural function and starts to do it well and independently.

This is the main new formation of the first psychological age and an expression of the crisis of the first year. In this case, the social situation of development should never be understood as a natural context of development, as is claimed in a majority of publications. We propose that the social situation of development might be understood technically as the actions of the adult toward the child at each period of development.

Cultural usage of objects should be differentiated from simple manipulation and exploration of objects and toys. Manipulation might be fulfilled separately by each hand, the child may move an object with no vocalization or without the presence and emotional support of an adult; there is no evidence of proper actions with an object, as the child may put it into their mouth,



FIGURE 2 | Manipulation with the toy (see text footnote 1).



FIGURE 3 | Action with the object (see text footnote 1).

drop an object, just simply touch it without no usage, and so on. In the case of appearance of proper cultural actions, the main differential features are as follows:

- 1 Eye movements and concentration of the glance toward an object.
- 2 Usage of both hands in coordinated movements trying to achieve proper actions with an object.
- 3 Intention of articulation of the words for this object or action.
- 4 Sharing with an adult or direction of an action with an object toward the adult with aspiration of approvement or agreement of the adult.

The difference between manipulation and actions with cultural objects directed to the established goal is essential for proper organization of a child's development during the crisis of the first year. Each adult has to give the child an opportunity to use a variety of cultural objects with the help, orientation, and oral explanation of the objects, actions, and situations. The adult continues to be representative of cultural experience and has to share this experience with the child. **Figure 2** shows manipulation, while **Figure 3** presents an example of the first cultural action in a child's life.

On the basis of such affirmation, we may confirm that the child takes part in the act of cultural communication. According to Eco (2005), the act of cultural communication begins when the participant can recognize an object of his/her communication. We believe that such a position might be useful for understanding not only anthropological and semiotic situations, but also the beginning of communication at the early stages of development. In the case of cultural development, the adult names the object to the child together while showing the proper action with the object. Later on, the child may recognize the object together with the verbal name and the corresponding action. Each action is introduced to the child by an adult, but the child must fulfill the action actively with the help of an adult. According to Leontiev (2009), the child's development is the acquisition of human actions, as a part of cultural history of humanity.

TABLE 4 | Content of the crisis of the first year.

Elements of the crisis	Indicators	Social situations
Intention for locomotion	Possibility to stand up and to make the first steps	Close presence of adult, who helps to realize the first steps
Intention for articulation of words	Possibilities to articulate syllables or simple words	Presence and affective communication of adult, who encourages pronunciation of the words together with the usage of objects
Intention for usage of cultural objects	Coordinate movements of both hands with the toys or objects	Presence of adult and presence of objects in practical situations of day-to-day life
Search for self-independence	Gestures, face expressions, crying with manifestation of necessity to be included into usage of new objects	Presence of adults who show interest in the child and approvement of his or her intentions

According to **Table 4**, it is possible to notice that the social situation of development might support, but also might be an obstacle for, the psychological development of the child. It is not enough to be with the child, but is also necessary to act in order to guarantee favorable psychological development. Assessment of the social situation of development might be helpful for elaboration of recommendations for parents and institutions for overcoming possible developmental difficulties (Solovieva and Quintanar, 2020). With the crisis of the first year, the next psychological period starts, in which systemic structure of the consciousness emerges; it happens while the meaning of historical and cultural objects becomes the meaning for the child (Vigotsky, 1984). **Table 4** includes the psychological content of the crisis of the first year.

DISCUSSION AND CONCLUSION

Psychological and neuropsychological literature commonly presents psychological development as synonymous to physical or physiological maturation, while psychological assessment of

a child's development is commonly reduced to assessment of movements, postures, and reflexes as progressive automatic changes during the first year (Gesell and Amatruda, 1980; Katona, 1988; Vojta, 2005; Bayley, 2015). In some proposals for assessment, mental development is reduced to natural progress in psychomotricity (Bayley, 2015). Frequently, different alterations of development are understood only as a result of brain damage or deficit of maturation, that is, are exposed as proper inner difficulties of the child and not as a result of the absence of joint activity between adult and child (Pelayo et al., 2016).

Vigotsky (1984) and his followers (Leontiev, 2000, 2003, 2009) have proposed another way of thinking. The child with or without difficulties is always a social child and is always a member of a social community. At the same time, psychology, in very few occasions, studies, what this community actually does with the child and how it guarantees psychological development. In this point our position, as positions of Vigotsky's followers, is radical: an adult's actions toward the child are the source of psychological development, but also the origin of the child's stagnation. The term of interaction is not enough for understanding the causes of development. Social interactions are everywhere in society. On the contrary, the term of activity is a precise term for what the adult does and what the child does. Interaction is a generic general word, while activity is a precise concrete term, which is useful for psychological research.

Some recent studies have shown the importance of development of actions with cultural objects during the following psychological age. In many cases, speech disturbances are related to the absence of the variety of proper cultural actions with objects. At the same time, gradual introduction and development of the possibility of usage of objects and toys serve as the basis for positive appearance of verbal expression and understanding in children with developmental difficulties (Borges, 2020; Borges et al., 2020a,b). Such importance helps us to consider in an original way the possibilities of the influence and correction of development in cases of difficulties or risks.

The concept of social situation of development, as proposed Vigotsky (1984), helps us to understand the guiding role of organized activity and of orientation for communication and manipulation during the first year of life. In posterior periods of development, methodological relation between assessment and correction of difficulties should take into account the social situation of development (Solovieva and Quintanar, 2020). Organization of psychological development should always lead to psychological development (Solovieva and Quintanar, 2014, 2015; Veraksa and Sheridan, 2018).

The article has presented the content of the first crisis as the result of gradual psychological development during the first psychological age. The crisis of the first year includes the appearance of the first words or intention for pronunciation of the words, which should be understood differently from initial communicative intention, which manifests in the animation complex of the first three months. In terms of Bozhovich (1981, p. 132), the central new psychological formation of the first

year is "affective representations" or "motivated representations." We may refer to it as affective and motivated representation of intention for communication. Another element of the content of the first year is aspiration for independent movement (walking) toward objects, including the adult. The third and central element of the crisis is appearance of cultural action with objects (spoon as the prototype). The actions of the child are not precise yet, but the intention of their action is very clear. The whole complex of their own intentions for words, walking, and actions with objects might be assumed as the major independence of the child. The child expresses unconscious necessity of usage of cultural objects independently. At the same time, the communicative goals of the child are conscious, he or she may already ask for external help and express their own need with gestures, movements, expressions, and importantly, with their own independent words, which may have autonomous phonetic structure and autonomous meaning (Vigotsky, 1982, 1984).

During the period of the crisis, the child is ready to change the social situation of development and the guiding activity through the acquisition of proper cultural actions with proper cultural meaning, mediatized by oral language as "psychological mean by excellency" (Vigotsky, 1982). Understanding the content of the first psychological age and the indicators of the crisis would serve as the basis for proper clinical and psychological assessment and for organization of the measures of corrections. The content of new psychological formations and their early or late appearance in each psychological age should become the objects of profound psychological and neuropsychological studies. "Probably, in [the] nearest future, neuropsychological exploration of cortical functions in newborn child[ren] and in early age would permit not only to precise deviations, but also to know how to eliminate them in correspondent time and how to provide individual programs for optimal development of the child" (Skvortsov, 1995, p. 93).

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

Written informed consent was obtained from the minor(s)' legal guardian/next of kin for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

The article is the result of the author's research and practice, dedicated to developmental psychology according to cultural historical conception of child's development. Both authors contributed to the article and approved the submitted version.

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Neuro-Behavioral Correlates of Executive Dysfunctions in Dyslexia Over Development From Childhood to Adulthood

Rola Farah^{1,2}, Silvio Ionta^{3,4} and Tzipi Horowitz-Kraus^{1,2*}

¹Educational Neuroimaging Center, Faculty of Education in Science and Technology, Technion, Haifa, Israel, ²Reading and Literacy Discovery Center and the Pediatric Neuroimaging Research Center, Cincinnati Children's Hospital Medical Center, Cincinnati, OH, United States, ³Sensory-Motor Lab (SeMoLa), Department of Ophthalmology, University of Lausanne, Lausanne, Switzerland, ⁴Jules Gonin Eye Hospital-Fondation Asile des Aveugles, Lausanne, Switzerland

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United States

*Correspondence:

Tzipi Horowitz-Kraus
tzipi.kraus@technion.ac.il

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Dyslexia is a neurobiological learning disability in the reading domain that has symptoms in early childhood and persists throughout life. Individuals with dyslexia experience difficulties in academia and cognitive and emotional challenges that can affect wellbeing. Early intervention is critical to minimize the long-term difficulties of these individuals. However, the behavioral and neural correlates which predict dyslexia are challenging to depict before reading is acquired. One of the precursors for language and reading acquisition is executive functions (EF). The present review aims to highlight the current atypicality found in individuals with dyslexia in the domain of EF using behavioral measures, brain mapping, functional connectivity, and diffusion tensor imaging along development. Individuals with dyslexia show EF abnormalities in both behavioral and neurobiological domains, starting in early childhood that persist into adulthood. EF impairment precedes reading disability, therefore adding an EF assessment to the neuropsychological testing is recommended for early intervention. EF training should also be considered for the most comprehensive outcomes.

Keywords: development, dyslexia, executive function, language, reading, neuroimaging

INTRODUCTION

Dyslexia: Definition and Characteristics

Since its first description over a century ago (Morgan, 1896), dyslexia has been investigated by cognitive and neurobiological studies to better understand the underlying mechanisms. One of the models that aim to describe the reading process in an attempt to explain the possible underlying impaired mechanisms in dyslexia is the simple view of reading model (Adlof et al., 2006). This model claims that reading comprehension is achieved by a combination of both language processing abilities and word decoding with EF abilities added to the model in recent years (Cutting et al., 2015). The present review summarizes the available neuro-behavioral evidence about such mechanisms to provide a compelling picture of dyslexia, the associated neuro-behavioral aspects, and possible input for intervention while focusing on the EF deficit in dyslexia.

Dyslexia is a neurobiological learning disability, affecting 5–17% of the population (Gabrieli, 2009) and is defined by word recognition difficulty and poor spelling abilities despite normal intelligence and adequate education and exposure to written material (Shaywitz and Shaywitz, 2008). Characteristics include inaccurate word recognition and decoding along with difficulties in reading comprehension (Directors of IDA, 2002). Word reading and reading comprehension have been found to be similar constructs; however, reading comprehension has been related to working memory abilities, and speed of processing was found to be a specific predictor for better word reading (Christopher et al., 2012).

Learning to read involves several critical steps: directing visual and auditory attention to the written stimuli and avoiding distractors, decoding of the word (i.e., phonological processing), visually perceiving the word and corresponding it to sound and receiving semantic information about the word (Horowitz-Kraus, 2016). Moving between the different steps demands mental resources crucial for successful reading, such as inhibition, working memory, shifting, and speed of processing (Booth et al., 2014). Therefore, it is not surprising that several theories have been raised to explain the underlying causes for reading deficits in dyslexia. The *phonological deficit theory* suggests that a dysfunction in the peri-sylvian region could lead to poor phonological skills in dyslexia (e.g., Ramus, 2003; Vellutino et al., 2004). This theory claims that the primary deficit in these readers might be the inability to translate written graphemes into their corresponding sounds due to a basic impairment in their phonological processor. An extension for this theory is called the “*double deficit theory*” suggesting a deficit not only in sound decoding and letter-sound matching but also in naming (letters, objects, etc.; Wolf and Bowers, 2000). The *orthographical deficit theory* postulates that dyslexia could stem from deficient orthographic imagery processing; these individuals suffer from an inability to perceive words holistically, leading to challenges in word recognition and the comprehension of orthographic information (Badian, 2005). This, in turn, leads them to an inability to establish a sufficient mental lexicon (i.e., self-teaching hypothesis; Share and Shalev, 2004). The *morphological deficit theory* suggests that dyslexia comes from poor knowledge of morphemes, decreasing written fluency (Nagy et al., 2006). The *asynchrony theory* (Breznitz, 2006) states that the core cause for dyslexia is a speed of processing deficit during word decoding. The *magnocellular deficit theory* has also been proposed, hypothesizing that the foundation of dyslexia arises from a dysfunction of the magnocellular visual system, causing a dysfunction in the processing of speedy temporal information (Stein and Walsh, 1997). The *cerebellar deficit theory* points at the dysfunction of the cerebellum in automatic word recognition (Nicolson et al., 2001). Lastly, a *temporal processing deficit theory* has been proposed, in that dyslexia would stem from a difficulty in fast temporal processing, most specifically in the low-level auditory domain (Tallal, 1980). More recently, the spread of modern neuro-investigation techniques to study dyslexia, the introduction of innovative experimental protocols, and the implementation of advanced data analyses brought to light the theory that the symptoms

of dyslexia could result from impaired executive functioning (EF; Helland and Asbjørnsen, 2000; Brosnan et al., 2002; Reiter et al., 2005; Berninger et al., 2006; Smith-Spark and Fisk, 2007; Altemeier et al., 2008; Horowitz-Kraus, 2014; Varvara et al., 2014; Butterfuss and Kendeou, 2017).

Executive Functions and Dyslexia

EFs is a broad term for top-down cognitive processes that aid in creating, planning, performing, and achieving goals (Lezak, 1982; Miyake et al., 2000). Three “main” EFs have been found *via* factor analyses: inhibition, shifting, and updating (Miyake et al., 2000) with others building upon Miyake and colleagues work, suggesting the inclusion of working memory and flexibility as well (Diamond, 2013). Inhibition is defined as the ability to impede automatic responses when necessary and can be tested by the Stroop (1935), anti-saccade (Hallett, 1978), and stop-signal tasks (Logan, 1994). Details regarding the sub-processes of inhibition during these paradigms can be found in Kok (1999). Shifting refers to switching between multiple tasks (Monsell, 1996) and can be assessed using the Wisconsin Card Sorting Task (Nyhus and Barceló, 2009), the Trail-Making Task Test B (Arbuthnott and Frank, 2000), and category switch test (Friedman et al., 2008). These functions are distinct yet rely on one another (Miyake et al., 2000). Other factor analyses on EFs and children point predominantly to working memory and shifting (Lehto et al., 2003; Huizinga et al., 2006) and processing speed (Span et al., 2004; Anderson and Reidy, 2012). A full list of tasks assessing different EF domains (i.e., updating, working memory, inhibition, shifting, short-term memory, and speed of processing) can be found in Friedman et al. (2008), Anderson and Reidy (2012), and Butterfuss and Kendeou (2017).

The development of EF is tightly connected to reading development. A theoretical review by Butterfuss and Kendeou (2017) demonstrated how EF is “embedded” within various reading models, including the construction-integration model (Kintsch, 1988), the structure-building framework (Gernsbacher, 1991), the resonance model (Albrecht and O’Brien, 1993), the event-indexing model (Zwaan et al., 1995), the casual network model (Trabasso et al., 1989), the constructionist theory (Graesser et al., 1994), and the landscape model (Van den Broek et al., 1999). The construction-integration model (Kintsch, 1988) involves related information links that assist each other and irrelevant information links that inhibit each other, relying on the EF inhibition. The structure-building framework (Gernsbacher, 1991) suppresses irrelevant information that does not correspond with the current structure. The resonance model (Albrecht and O’Brien, 1993) found that phrases related to the text strengthened the target, whereas irrelevant phrases were suppressed. The event-indexing model (Zwaan et al., 1995) involves shifting from one dimension (i.e., time, space, causality, motivation, and agents) to another. The casual network model (Trabasso et al., 1989) includes memory for narrative elements (i.e., settings, events, goals, attempts, outcomes, and reactions). The constructionist theory (Graesser et al., 1994) contains cognitive control mechanisms in the search for semantics, likely shifting. Even higher-level EFs, such as planning, have also

been hypothesized to facilitate reading (Kendeou et al., 2015). These models highlight that EF plays a crucial role in reading. However, it is important to note that not one single model can fully explain the heterogeneous patterns in both reading and EF abilities found in individuals with dyslexia.

Unlike language development abilities that reach their peak in early childhood (age 7; Purves et al., 2001), EF may mature during adulthood (mid-twenties; Romine and Reynolds, 2005). Therefore, to better understand how academic achievements, especially reading, rely on EF, it is important to discuss the time each of these abilities develops in life. Some components of EF develop relatively early along development and some mature later on until a full maturation of the prefrontal cortex at the age of 25 (Giedd et al., 2009). Inhibition has been shown to develop at 12 months (Diamond and Goldman-Rakic, 1989), reaching adult levels at age 12 years (Welsh et al., 1991). Planning skills have been found to fully mature between 9 to 13 years old (Welsh et al., 1991; Anderson et al., 1996). Shifting begins to occur around 4 to 5 years (Espy, 1997) and increases significantly at 7 years old (Anderson, 2002) and updating, which requires monitoring and coding of information in memory and is related to verbal and visuospatial working memory abilities (St Clair-Thompson and Gathercole, 2005). Therefore, not all EFs may follow the same developmental trend (Passler et al., 1985; Lehto et al., 2003; Jurado and Rosselli, 2007), and some studies have found later ages of EF maturation (Huizinga et al., 2006) or that processing speed could be the driving factor of EF maturation in children (Span et al., 2004). A longitudinal study done by Altemeier et al. (2008) found that all EFs may not develop the same in typical readers grades 1 to 6: inhibition abilities can increase consistently, whereas rapid automatic switching and combined inhibition and switching may begin to slow in the rate along development around fourth grade. However, another theory suggested by Miyake and Friedman (the unity/diversity framework) claims that EF (including updating, shifting, and inhibition) is relatively stable along development (Miyake and Friedman, 2012).

In sum, although reading acquisition is formally acquired at the age of 6 and EF fully matures later in life, these abilities are tangled in the reading process. A crucial question arises as to the involvement of EF in the atypical course of reading development (i.e., in dyslexia), using behavioral and neurobiological measures. Of note, dyslexia often co-occurs with attention deficit hyperactivity disorder, comorbidity characterized by EF deficit (Pennington et al., 1993; Willcutt et al., 2001, 2005). However, as the current review does not focus on comorbidities, it includes studies focusing only on dyslexia and the involvement of EF in this disorder, focusing on the developmental model of sub-components of EF, based on Anderson's approach (Anderson and Reidy, 2012). This model was chosen as it relates to the development of several sub-components of EF (not limited to the three core components), as well as to the attention system from birth onwards, which provides a longitudinal framework to this review focusing on the involvement of EF in dyslexia along life span.

MATERIALS AND METHODS

Searching the Relevant Papers

PubMed and Google Scholar were used to search for studies exploring behavioral and neurobiological dysfunction in dyslexia. The following keywords were used as: "executive function," "cognitive control," "functional MRI," "structural MRI," "EEG," "reading," and "dyslexia" as well as the combinations including the cognitive abilities/dyslexia and neuroimaging methods. This search generated over 200,000 manuscripts with 171,000 manuscripts for executive functions/cognitive control and "dyslexia" and approximately 38,000 for "dyslexia" and functional/structural "MRI" or "EEG." The studies reporting of evidence about neurobiological changes in dyslexia in relation to EF were included in this nonsystematic review. The brain regions per developmental group were defined based on the automated anatomical labeling (AAL) atlas (Tzourio-Mazoyer et al., 2002). AAL is a software commonly implemented within neuroimaging analysis tools, such as Statistical Parametric Mapping to identify the brain regions comprised within specific neural activation blobs according to a standard brain atlas (for more information see Rolls et al., 2020). All images were created using the BrainNet Viewer (Xia et al., 2013).

RESULTS

Individuals with dyslexia demonstrate deficits in EFs, with a varied profile along development (see **Table 1**). Children at risk for dyslexia may demonstrate more challenges in selective attention and visuospatial short-term memory. Children show more deficits in planning, teenagers show more deficits in speed of processing, and adults show difficulties in planning and speed of processing. All age groups show deficits in working memory. One possibility for these changes along age is the gradual maturation and increased connections within the frontal lobe and between the frontal lobe and other brain regions (Fair et al., 2009).

Behavioral Evidence

Executive Dysfunctions in Individuals With Dyslexia

EFs develop along life span (Diamond, 1985; Altemeier et al., 2008). Additionally, in individuals with reading difficulties, some longitudinal studies reported deficits in working memory observed from age 6 to 49 years (Chiappe et al., 2000). As explained, due to brain maturation differences, especially in relatedness to EF, the deficit in EFs among individuals with dyslexia in 3 age groups will be reviewed: children "at-risk" for dyslexia (ages 0–5 years), children (ages 6–12 years), adolescents (ages 13–21), and adults (ages 22 and up), both behaviorally and neurobiologically.

Executive Dysfunctions in Children at Risk for Dyslexia (0–5 Years)

Children at risk for dyslexia (i.e., with parents or siblings with dyslexia), between the ages of 3 to 5 years, show EF impairment,

TABLE 1 | Executive dysfunctions in individuals with dyslexia along development.

Executive function	Children at risk (0–5)	Children with dyslexia (6–12)	Teenagers with dyslexia (13–21)	Adults with dyslexia (22+)
Inhibition	Gooch et al., 2014	Lazarus et al., 1984; Kelly et al., 1989; Everatt et al., 1997; Helland and Asbjørnsen, 2000; Carretti et al., 2005; Reiter et al., 2005		
Memory	Gooch et al., 2014	Fein et al., 1988; Barnea et al., 1994; Chiappe et al., 2000; Brosnan et al., 2002; Reiter et al., 2005; Berninger et al., 2006	Fein et al., 1988; Chiappe et al., 2000; Brosnan et al., 2002; Carretti et al., 2005; Horowitz-Kraus, 2015	Fein et al., 1988; Chiappe et al., 2000; Brosnan et al., 2002; Berninger et al., 2006; Horowitz-Kraus and Breznitz, 2009
Shifting		Kelly et al., 1989; Kershner and Morton, 1990; Helland and Asbjørnsen, 2000; Horowitz-Kraus, 2014	Asbjørnsen and Bryden, 1998; Kraus and Horowitz-Kraus, 2014; Horowitz-Kraus, 2015	
Speed of processing			Horowitz-Kraus, 2015	Breznitz and Misra, 2003; Horowitz-Kraus and Breznitz, 2011
Attention	Facoetti et al., 2010; Gooch et al., 2014; Hardeman, 2016	Facoetti et al., 2000		
Problem solving and planning		Chiarenza, 1990; Levin, 1990; Mati-Zissi et al., 1998; Reiter et al., 2005		Catts, 1989; Weyandt et al., 1998

in the domains of selective and sustained attention, inhibition, and visuospatial short-term memory which were found to be correlated with language ability (Gooch et al., 2014). Executive functions play a role as a predictor of future reading disability at 4.5 years old in a population at risk (Thompson et al., 2015). Three-year-olds at risk of dyslexia exhibited a trend of lower scores on selective attention. However, the effects of inhibition or working memory were not significant (Hardeman, 2016). Overall, the literature related to children at risk for dyslexia before reading age points at attention, inhibition, and visuospatial short-term memory as EF components which may be “precursors” for dyslexia before reading age.

Executive Dysfunctions in Children With Dyslexia (6–12 Years)

Children with dyslexia (6–12 years old) have shown impairment in inhibition, as assessed by the Stroop task (Lazarus et al., 1984; Kelly et al., 1989; Everatt et al., 1997; Helland and Asbjørnsen, 2000; Reiter et al., 2005), as well as in verbal and nonverbal working memory (Fein et al., 1988; Barnea et al., 1994; Brosnan et al., 2002; Reiter et al., 2005; Berninger et al., 2006). Children with dyslexia ages 6–12 years also show difficulties with planning (Chiarenza, 1990; Levin, 1990; Mati-Zissi et al., 1998), as shown by worse performance on the Tower of London (Reiter et al., 2005). Children with dyslexia ages 11 and 12 display complications on shifting (Kelly et al., 1989; Kershner and Morton, 1990; Helland and Asbjørnsen, 2000), also shown through more errors and a slower reaction time when performing the Wisconsin Card Sorting Test (Horowitz-Kraus, 2014). Carretti et al. (2005) found that poor readers made more intrusion errors, supporting that working

memory may aid reading comprehension through inhibition in children. Related to the visual attention difficulties suggested in adults (Smith-Spark and Fisk, 2007), 8–17-year-old children with reading difficulties showed decreased visual and auditory spatial attention difficulties which were also related to their decreased reading abilities (Varvara et al., 2014). By that, the authors concluded that these readers showed a deficiency in their central executive system (Varvara et al., 2014). These results were also observed by others and extended to switching/shifting abilities as measured using the Wisconsin task in children ages 8–17 years old with dyslexia (Menghini et al., 2010). Overall, research on beginning readers points to inhibition, visual/auditory attention, and working memory dysfunction, along with planning and shifting challenges in 6–12-year-old children as altered EF in children with dyslexia compared to age-matched typical readers.

Executive Dysfunction in Teenagers With Dyslexia (Ages 13–21 Years)

Teenagers with dyslexia ages 13–21 years old exhibited deficits in EFs, specifically in the domains of verbal and nonverbal working memory (Fein et al., 1988; Brosnan et al., 2002; Horowitz-Kraus, 2015), shifting (Asbjørnsen and Bryden, 1998), and speed of processing (Horowitz-Kraus, 2015). In other studies, readers with dyslexia also showed deficits in error detection during the Madrid Card Sorting Task, similar to the Wisconsin Card Sorting Task assessing shifting/switching, with slower reaction times and more errors (Kraus and Horowitz-Kraus, 2014; Horowitz-Kraus, 2015). These readers also showed impairment in error monitoring in reading tasks as well (Horowitz-Kraus, 2011). In general, research highlights difficulties

both in more basic EF as well as more complex EFs, such as in the domains of working memory, shifting, speed of processing, and also error detection and monitoring for teenagers with dyslexia.

Executive Dysfunction in Adults With Dyslexia (22 Years and Older)

Adults with dyslexia have shown difficulties with planning (Catts, 1989; Weyandt et al., 1998), working memory (Fein et al., 1988; Brosnan et al., 2002; Berninger et al., 2006; Horowitz-Kraus and Breznitz, 2009), and visual processing (visual-spatial working memory) abilities (Smith-Spark et al., 2016; Provazza et al., 2019). College students with dyslexia have shown impairments in speed of processing (Breznitz and Misra, 2003; Horowitz-Kraus and Breznitz, 2011), which were related to their reading accuracy and reaction time. Brosnan's significant differences between adults with dyslexia and typical readers were found in the EF domains of planning, sequencing, and organization of memory and visual-spatial measures (Brosnan et al., 2002). However, Smith-Spark and colleagues suggested that the difficulties in working memory were extended into visual-spatial attention abilities in adulthood, which support a central difficulty in EF in this population (Smith-Spark et al., 2003; Smith-Spark and Fisk, 2007). These findings were echoed by Provazza and colleagues demonstrating a similar visual processing challenge in this population (Provazza et al., 2019). Considering the brain networks involved, for example, in visuospatial processing (Perruchoud et al., 2016) or attention (Pamplona et al., 2020) comprise a large number of brain regions and related interconnections both in adulthood and development (Ionta, 2021), it is possible that in individuals with dyslexia, possibly over time/age, natural neuroplastic compensatory mechanisms are put in place to establish alternative neural activations/connections which would make dyslexic people able to compensate their deficits and resemble the performance of their age-matched non-dyslexic peers in the domains of organization, visual-spatial abilities, shifting, and attention.

Despite the great benefit behavioral and cognitive testing provide when discussing reading and EF abilities in those with dyslexia, one limitation of behavioral tests is that they can be considered versatile in the functions they assess. As there is a current debate in the literature of the definition and assessment of EFs, the current strength of validity in EF assessment should be taken with some caution (Jurado and Rosselli, 2007). However, the behavioral tests used in this review have been used for many years and in many studies in assessing EFs (Axelrod et al., 1994; Romine et al., 2004; Carlson, 2005). Neuroimaging data can assist with this limitation, by giving the ability to differentiate network functionality. As different networks have been attributed to more basic attention abilities and to higher-level monitoring, different aspects of EFs can be separated and assessed using neuroimaging techniques (Dosenbach et al., 2008; Petersen and Posner, 2012).

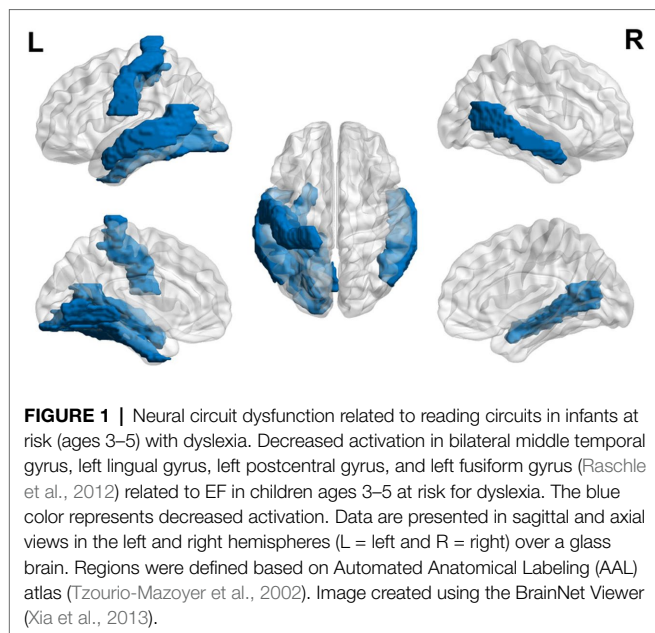
The Neurobiology of Executive Function in Dyslexia

EFs seem to be important in the reading process, as EF areas (i.e., the inferior frontal gyrus, middle frontal gyrus, precuneus, and posterior cingulate) are also involved during reading-related tasks in individuals from 5 to 18 years (Karunanayaka et al., 2007) as well as in a listening-doing matching system (Halje et al., 2015). Additionally, since EFs rely on multiple brain regions, they may be particularly sensitive to brain dysfunction (Reiter et al., 2005). Brain regions associated with EFs have also been found to be correlated with reading ability. Greater functional connectivity of an EF network (i.e., the cingulo-opercular network) has been found, accompanying gains in both reading and EF behavioral measures after a reading intervention (Horowitz-Kraus et al., 2015). Greater connectivity between EF and visual regions has also been correlated with greater reading comprehension post-reading intervention (Horowitz-Kraus et al., 2014). Overall, the scientific literature provides evidence for the importance of neurobiological EF measures for reading. Here, we review the neurobiology of dyslexia in a developmental manner, focusing on EF networks during reading-related tasks.

Neural Circuits Related to EF in Children at Risk for Dyslexia (0–5 Years)

Most of the current neuroimaging research done with infants and young children at risk for dyslexia is with electroencephalography (EEG) and event-related potentials (ERPs). When compared to the behavioral performance of a head-turn task, smaller bilateral response in at risk infants was associated with greater performance, whereas in controls, greater left hemisphere response was associated with greater performance, suggesting that at risk infants may have differential neural processes involved for auditory and language tasks already at 6 months (Lyytinen et al., 2004). Zuijzen et al. (2013) found that 2-month-olds at risk for dyslexia who later at age 7 years scored poorly on a word reading fluency measure did not show a mismatch response, whereas infants at risk for dyslexia and controls who later performed well on a fluency measure did show a mismatch response, showing differentiation of processing to two different auditory sounds (Zuijzen et al., 2013). These aberrant event-related responses are also found in 6-month-olds at risk for dyslexia (Leppänen et al., 2002) and *via* delayed P100 [representing selective attention (Mangun and Hillyard, 1991)] and P200 [associated with working memory and attention (Lijffijt et al., 2009)] peaks for standard auditory stimuli in children at risk for dyslexia (van Herten et al., 2008). The N200 response [an ERP related to inhibition (Heil et al., 2000)] is absent in at risk 2-year-olds compared to controls during lexical-semantic priming (von Koss Torkildsen et al., 2007).

Using functional MRI in 5-year-old children at risk for dyslexia, children were asked to listen to two words and decide if they both started with the same beginning sound (Raschle et al., 2012). Children at risk for dyslexia exhibited hypoactivation in bilateral occipito-temporal and left temporo-parietal regions (Raschle et al., 2012; see **Figure 1**). This finding corresponds



to a decrease in gray matter found in the left occipito-temporal, bilateral parieto-temporal, left fusiform gyrus, and right lingual gyrus (Raschle et al., 2011). In as young as 6 to 18 months, Langer et al. (2017) found lower fractional anisotropy in the left arcuate fasciculus. Children ages 3–5 years showed increased functional connectivity of their future reading network and language processing regions and regions influencing EFs (i.e., left Brodmann area 2, 13, and 44 and right Brodmann area 6 and 44) for greater maternal fluency ability (Horowitz-Kraus et al., 2017).

Neural Circuits Related to EF in Children With Dyslexia (6–12 Years)

Using EEG, Duffy et al. (1980) found higher alpha values in children with dyslexia in the bilateral medial frontal region during a naming abstract figures task and a reading task and only in the left medial frontal region during speech, a sound-symbol-association test, and during rest with eyes open (Duffy et al., 1980). Higher alpha values were also observed during a Kimura figures test (assessing nonverbal memory) in the left anterolateral frontal region (Duffy et al., 1980). The authors suggest this may represent a hypoactivation of frontal systems (Duffy et al., 1980), conflicting with the hyperactivation seen in frontal networks in fMRI studies.

Some studies have shown that children with dyslexia may display neurobiological dysfunction related to EF as well as language systems during reading tasks. During a narrative comprehension task, hyperactivation in the right superior frontal gyrus and right middle frontal gyrus, both areas involved in EF, was observed in children with dyslexia (Horowitz-Kraus et al., 2016). During sentence comprehension, children with dyslexia exhibited hyperactivation in the left middle/superior temporal gyri and bilateral insula [part of the cingulo-opercular network (Power et al., 2011)], right cingulate gyrus [also in

the cingulo-opercular network (Power et al., 2011)], right superior frontal gyrus [an area involved in working memory tasks (Johnson et al., 2003)], and the right parietal lobe (Rimrodt et al., 2008). During a reading task, children with dyslexia displayed hypoactivation in phonological areas, such as the left fusiform gyrus (visual word form area) and Wernicke's area, whereas hyperactivation was seen in bilateral orthographic areas (i.e., anterior visual word form areas and posterior bilateral middle temporal gyri; Saralegui et al., 2014). The authors hypothesize that individuals with dyslexia compensate for phonological deficits by hyper activating areas in the orthographic route.

Seki et al. (2001) also found compensatory activation during a reading task in the bilateral occipital cortex, inferior frontal regions (areas involved in EF), and inferior precentral gyrus. The involvement of visual and EF-related regions was also observed during a phonological task, one of the basic abilities impaired in dyslexia: hyperactivation of the left inferior frontal gyrus (an area involved in EF; Georgiewa et al., 2002) and hypoactivation of the right visual and left occipital cortex (Shaywitz et al., 2002; Liu et al., 2012) and temporal and prefrontal cortices (involved in EF; Backes et al., 2002; Shaywitz et al., 2002), specifically the left superior temporal gyrus (Kita et al., 2013), the left fusiform cortex (Desroches et al., 2010), and left inferior frontal gyrus (an area involved in EF; Liu et al., 2012).

Interestingly, also subcortical regions were found to show hypoactivation during a phonological processing task in children with dyslexia, such as the basal ganglia (Kita et al., 2013), in addition to the left extrastriate cortex (Backes et al., 2002) and the right cerebellum (van Ermingen-Marbach et al., 2013). Considering the importance of mutual exchanges between cortical and subcortical regions (Zeugin and Ionta, 2021), these findings demonstrate how more basic learning mechanisms related to cortico-subcortical-cerebellar activations are also different in children with dyslexia. It has been proposed that this hyperactivation in the above-mentioned EF regions and hypoactivation of visual processing and language-related regions is related to the pathology of dyslexia, through a greater attempt of recognizing words holistically and retrieving the semantic meaning of it from working memory and allocating greater attention for error detection to compensate for hypoactivation in visual and reading-related areas.

Shaywitz et al. (2002) found greater activation in bilateral inferior frontal gyri (areas involved in EF) in older children. These results can be viewed in a network-based framework: higher global efficiency in the fronto-parietal network was negatively correlated with cognitive tests on narrative comprehension, phonological awareness, word and non-word reading, and executive abilities (assessed *via* the Stroop task; Horowitz-Kraus et al., 2016). When analyzed longitudinally, an upregulation of connectivity in occipito-temporal connections and a downregulation in inferior frontal gyri connections was found from 6 to 8 years, but from 8 to 12 years connectivity was similar to controls. This finding supports the hypothesis that abnormalities in the EF network could precede dysfunction

in the reading network (Clark et al., 2014). Overall, increased right hemisphere connectivity has been found in children with dyslexia (Finn et al., 2014).

Extending the functional changes in EF and visual related regions also to the structural domain in children with dyslexia in this age group was suggested by Williams et al. (2017) who found thinner cortex in bilateral occipito-parietal and inferior temporal cortices (i.e., portions of the reading network) compared to controls (Williams et al., 2017). Thinner cortex was also found in areas relevant for cognitive control (i.e., the right orbitofrontal, left anterior cingulate, left superior parietal, and right medial parietal cortices). The orbitofrontal cortex is involved in learning from probabilistic feedback (Tsuchida et al., 2010) and in decision making (Kringelbach, 2005). This finding could explain why lower error-related negativities (i.e., a neural mechanism that is activated when an error is made) and a higher error rate have been found in dyslexics compared to controls (Horowitz-Kraus, 2012). Support for the structural alterations in EF and visual regions in children with dyslexia is provided by diffusion tensor imaging studies. Slight fractional anisotropy (FA) decreases have also been found in left temporo-parietal neural pathways in children with dyslexia (Deutsch et al., 2005). Further, children with dyslexia showed reduced FA in the left superior longitudinal fasciculus, connecting the frontal and parietal lobes (Carter et al., 2009; Rollins et al., 2009), along with the left corona radiata, the left centrum semiovale (Odegard et al., 2009), the left inferior frontal gyrus (an area involved in EF), and temporo-parietal areas (Rimrodt et al., 2010). Corpus callosum differences are also seen, with a smaller genu that runs along the anterior cingulate cortex (related to EF) in dyslexic children (Hynd et al., 1995).

Overall, the literature suggests that children with dyslexia ages 6 to 12 years demonstrate hyperactivation in areas related to EF and hypoactivation in areas related to language and visual areas during reading-related tasks using MRI (see Figure 2). Nine- and 10-year-olds demonstrate increased alpha in frontal areas during reading-related tasks. Structural differences are also found with the thinner cortex in reading-related and EF areas.

Neural Circuits Related to EF in Teenagers With Dyslexia (13–21 Years)

As individuals with dyslexia reach teenage years, dysfunction has been found in EF circuits during reading tasks as well, with reports showing changes in brain responses during the Wisconsin shifting/swathing task (decreased feedback-related negativity, i.e., an ERP related to brain activation associated with a response to feedback and is part the monitoring system) in children ages 12–14 years with dyslexia vs. typical readers (Kraus and Horowitz-Kraus, 2014).

When assessing teenagers with dyslexia versus age- and reading-matched typical readers during a visual word rhyme judgment task (assessing the phonological analysis of orthographic input), hyperactivation was found in EF areas

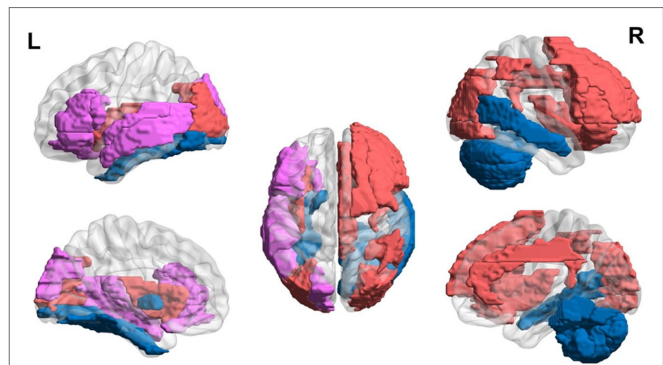


FIGURE 2 | Neural circuit dysfunction related to reading circuits in children (ages 6–12) with dyslexia. Decreased activation in left superior temporal gyrus (Kita et al., 2013; Saralegui et al., 2014), bilateral middle temporal gyrus (Saralegui et al., 2014), left fusiform gyrus (Desroches et al., 2010; Saralegui et al., 2014), left globus pallidus (Kita et al., 2013), left inferior frontal gyrus (Liu et al., 2012), right cerebellum (van Ermingen-Marbach et al., 2013), and left superior occipital gyrus (Backes et al., 2002; Shaywitz et al., 2002) and increased activation in the right superior frontal gyrus (Horowitz-Kraus et al., 2016), right middle frontal gyrus (Horowitz-Kraus et al., 2016), bilateral insula (Rimrodt et al., 2008), left superior temporal gyrus (Rimrodt et al., 2008), left middle temporal gyrus (Rimrodt et al., 2008), right cingulum (Rimrodt et al., 2008), right inferior parietal lobule (Rimrodt et al., 2008), bilateral occipital gyrus (Seki et al., 2001), and bilateral inferior frontal gyrus (Seki et al., 2001; Georgiewa et al., 2002; Shaywitz et al., 2002), related to EF in children ages 6–12 at risk for dyslexia. Red represents areas of hyperactivity, blue areas of hypoactivity, and purple areas where papers have reported hyperactivity and hypoactivity. Data are presented in sagittal and axial views in the left and right hemispheres (L = left and R = right) over a glass brain. Regions were defined based on AAL atlas (Tzourio-Mazoyer et al., 2002). Image created using the BrainNet Viewer (Xia et al., 2013).

(i.e., the left inferior and middle frontal gyri and caudate) and the thalamus compared to age-matched controls (Hoeft et al., 2007). However, additional support for alteration related to visual processing was observed by hypoactivation in left parietal and fusiform regions with both age- and reading-matched controls (Hoeft et al., 2007). However, hyperactivation has also been found in visual areas (Wimmer et al., 2010). Hypoactivation seen in dyslexia could then be attributed to a malfunction in “classic” dyslexic posterior malfunction, whereas the hyperactivation could be a compensatory response in EF areas to offset the dysfunction found in posterior areas (Hoeft et al., 2007). Desroches et al. (2010) also found hypoactivation of the left fusiform cortex during a phonology task. During a phonological task, hypoactivation in the posterior regions of the reading network (Brambati et al., 2006), in a left ventral occipito-temporal region, a left inferior parietal region, and a left inferior frontal region [an area related to EF (Wimmer et al., 2010; Steinbrink et al., 2012)] was observed. During an N-back task, children with dyslexia displayed hypoactivation in the left superior parietal lobule and the right inferior prefrontal gyrus (Beneventi et al., 2010). During the Madrid Card Sorting Task, teenagers with dyslexia showed decreased target-locked N100, involved in selective attention (Hillyard et al., 1973), and P300, also involved in attention (Polich, 1986) and amplitudes (Kraus and Horowitz-Kraus, 2014; Horowitz-Kraus, 2015).

An altered participation of EF during phonological processing was observed *via* functional connectivity between phonological processing-related regions and an EF network (Wolf et al., 2010). A left prefrontal network exhibited increased connectivity in the left prefrontal and inferior parietal regions (Wolf et al., 2010). A bilateral executive fronto-parietal network showed decreased connectivity in bilateral dorsolateral prefrontal and posterior parietal regions and increased connectivity in the left angular gyrus, left hippocampus, and right thalamus (Wolf et al., 2010). Weakened connectivity in the left fronto-parietal network in dyslexia, even after behavioral remediation, has been shown (Koyama et al., 2013). This further represents the dysfunction in both EF and reading networks in teenagers with dyslexia. Hyperactivation of the right inferior frontal gyrus in teenagers with dyslexia during a reading task predicted greater reading improvement 2 years later, showing that possibly greater activation in frontal areas contributes to better gain later on (Hoeft et al., 2007).

In sum, teenagers with dyslexia (ages 13–21 years) exhibit hyperactivation in EF areas (i.e., left inferior and middle frontal gyri and caudate) and the thalamus and hypoactivation in visual areas (i.e., left parietal and fusiform regions) and in posterior regions of the reading network. However, conflicting results of hypoactivation in the left and right inferior frontal region and hyperactivation in visual areas have been found as well. A general trend of hyperactivation in EF areas and hypoactivation in reading areas is found, but more studies need to be done to clarify conflicting results (Figure 3).

Neural Circuits Related to EF in Adults With Dyslexia (22 Years and Older)

Lastly, studies have shown that adults with dyslexia may continue showing executive dysfunction during reading-related tasks (Figure 4). College students with dyslexia have been found to have lower error-related negativity amplitudes and later latencies in error responses compared to controls (Horowitz-Kraus and Breznitz, 2008, 2009).

Reading with time constraints shows neurobiological differences in regions related to vision and EF in adults with dyslexia (Karni et al., 2005). During fast reading, no significant activation differences were seen in the two groups. However, during slow non-word reading, hyperactivation in the left inferior frontal gyrus (involved in EF) and operculum was found, compared to the controls that showed activation in the visual areas (Karni et al., 2005). During a phonological task, hypoactivation in the auditory sensory thalamus (i.e., the medial geniculate body; Díaz et al., 2012), in a left ventral occipito-temporal region (related to orthography), a left inferior parietal region (related to attention) and a left inferior frontal region (Wimmer et al., 2010), and hyperactivation in visual occipital regions (Wimmer et al., 2010), and asymmetry in the inferior frontal gyrus (an EF area; Hernandez et al., 2013) was found in adults with dyslexia compared to controls.

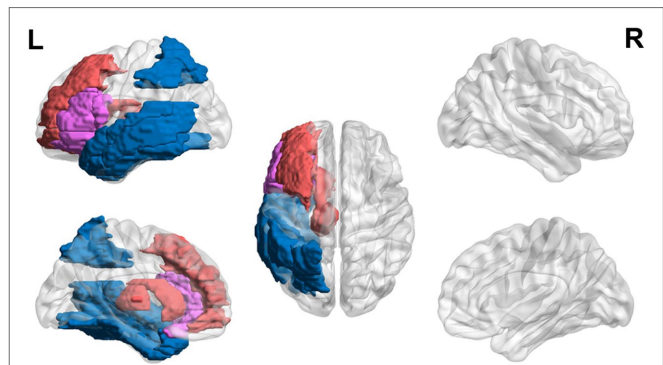


FIGURE 3 | Neural circuit dysfunction related to reading circuits in teenagers (ages 13–21) with dyslexia. Decreased activation in the left inferior frontal gyrus (Wimmer et al., 2010; Steinbrink et al., 2012), left parietal lobule (Hoeft et al., 2007; Wimmer et al., 2010; Steinbrink et al., 2012), left fusiform gyrus (Hoeft et al., 2007; Desroches et al., 2010), and left temporal gyrus (Wimmer et al., 2010; Steinbrink et al., 2012) and increased activation in the left inferior frontal gyrus (Hoeft et al., 2007), left middle frontal gyrus (Hoeft et al., 2007), left caudate (Hoeft et al., 2007), and left thalamus (Hoeft et al., 2007) related to EF in children ages 13–21 at risk for dyslexia. Red represents areas of hyperactivity, blue areas of hypoactivity, and purple areas where papers have reported hyperactivity and hypoactivity. Data are presented in sagittal and axial views in the left and right hemispheres (L = left and R = right) over a glass brain. Regions were defined based on AAL atlas (Tzourio-Mazoyer et al., 2002). Image created using the BrainNet Viewer (Xia et al., 2013).

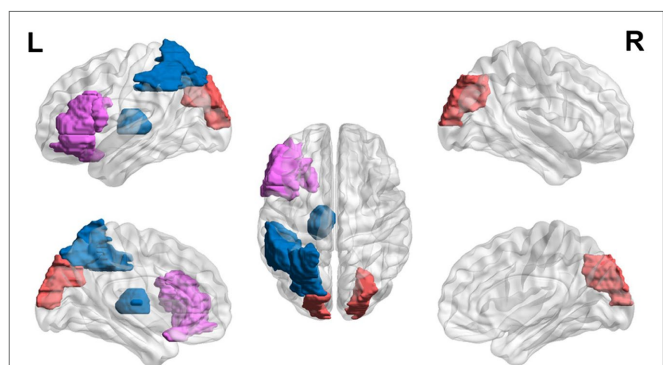


FIGURE 4 | Neural circuit dysfunction related to reading circuits in adults (ages 22+) with dyslexia. Decreased activation in the left thalamus (Díaz et al., 2012), the left inferior frontal gyrus (Wimmer et al., 2010), and the left parietal lobule (Wimmer et al., 2010) and increased activation in the left inferior frontal gyrus (Karni et al., 2005) and bilateral superior occipital gyri (Wimmer et al., 2010) related to EF in adults ages 21 and up with dyslexia. Red represents areas of hyperactivity, blue areas of hypoactivity, and purple areas where papers have reported hyperactivity and hypoactivity. Data are presented in sagittal and axial views in the left and right hemispheres (L = left and R = right) over a glass brain. Regions were defined based on AAL atlas (Tzourio-Mazoyer et al., 2002). Image created using the BrainNet Viewer (Xia et al., 2013).

Further, structural data supported these abnormalities in both EF and reading networks. Hinting at the importance of multisensory processing (Zeugin et al., 2017), decreased FA has been found in bilateral fronto-temporal and left

temporo-parietal white matter regions (Steinbrink et al., 2008) and left middle and inferior temporal gyri and left arcuate fasciculus (Silani et al., 2005; Vandermosten et al., 2012). Decreased FA was found in bilateral temporo-parietal white matter regions in another sample of adults (Klingberg et al., 2000), and a decrease in FA in all four lobes has been seen (Richards et al., 2008).

Overall, the research supports that individuals with dyslexia may display difficulties in error detection and hypoactivation in reading-related and EF areas and hyperactivation in visual areas during phonological tasks even when reaching adulthood (ages 21 and up).

DISCUSSION

All age groups show dysfunction in neural circuits related to EF, showing that EF is involved in reading tasks and individuals with dyslexia experience a malfunction in these areas. Although the studies in the present review outline the abnormal activity of neural circuits related to EF and reading, another alternative is that abnormalities in brain areas related to EF could reflect compensation effects. While assessing very young children to determine the core neurobiological function of dyslexia are difficult, some studies have been done to determine when frontal dysfunction begins in children at risk for dyslexia. Delayed P100 and P200 peaks were seen during auditory stimulus presentation in 17-month-olds at risk for dyslexia (van Herten et al., 2008) and 3- to 5-year-old children with greater maternal fluency ability show increased functional connectivity between future reading networks and EF-related regions (Horowitz-Kraus et al., 2017), solidifying that EFs are developing quite early in correspondence with reading networks.

Overall, this review suggests a consistent phenomenon of lower behavioral EF abilities and alteration of neural circuits related to EF along development. These alterations were found in functional, structural, and network measures generated from MRI data as well as from EEG. These differences were also related to changes in visual processing regions [i.e., a left ventral occipito-temporal region, extrastriate regions, left parietal and fusiform regions, and the right and left occipital cortex (Shaywitz et al., 2002; Karni et al., 2005; Hoeft et al., 2007; Wimmer et al., 2010; Liu et al., 2012)].

One explanation of how EFs and reading development are connected can be explained by the inside-out and outside-in model. Emergent literacy involves both inside-out skills, such as phonological awareness and outside-in skills, such as conceptual knowledge (Whitehurst and Lonigan, 1998). EF is positioned in this model both as an inside-out factor which is part of the child's essential abilities to learn language and reading and is involved in the outside-in factors contributing to reading by allowing the child to attend to stories, books, and other literacy material provided in the child's development (Whitehurst et al., 1988). These data support the tight relations between nature (inside-out abilities) and nurture (outside-in factors, such as exposure to language, literacy, and parental reading) on neural circuits supporting future reading in the

developing brain. See **Table 2** for an overview of the neural circuits related to EF along development in children at risk and with dyslexia as well as in adults.

Limitations

Some limitations exist in exploring if EF deficits in dyslexia occur before dysfunction in reading. It is important to note that some studies exploring EF and reading comprehension have found no correlation between the two, or only between certain EFs (Pennington et al., 1993; Nydén et al., 1999; Willcutt et al., 2001, 2005, 2010; Jeffries and Everatt, 2004; Cain, 2006; Bental and Tirosh, 2007; Altemeier et al., 2008; Tiffin-Richards et al., 2008; de Jong et al., 2009; Menghini et al., 2010; Gooch et al., 2011; Christopher et al., 2012; Peng et al., 2013; Varvara et al., 2014; Moura et al., 2015, 2017; Wang and Yang, 2015). However, the inconsistency in the research has been hypothesized to be due to several factors, including group classification difficulties, theoretical definition inconsistencies, and task impurity (Doyle et al., 2018). Lastly, although some developmental theories agree that speed of processing abilities is part of EF (Anderson and Reidy, 2012), this view is still under debate in the scientific community (Gordon et al., 2020).

Conclusion

This review aimed to highlight the current behavioral and neurobiological atypicalities found in dyslexics along development in reading, highlighting EF regions and networks. As outlined earlier, individuals with dyslexia show altered brain activation and lower performance in higher-order EF tasks (such as WCST; Horowitz-Kraus, 2014, 2015; Kraus and Horowitz-Kraus, 2014), tasks which also involve reading abilities (e.g., Stroop task; Lazarus et al., 1984; Kelly et al., 1989; Everatt et al., 1997; Helland and Asbjørnsen, 2000; Reiter et al., 2005), or tasks that involve an auditory component (e.g., mismatch negativity; Lyytinen et al., 2004; van Herten et al., 2008; Raschle et al., 2012; Zuijlen et al., 2013). One can postulate that one of the reasons for this altered performance is the tasks' complexity, the involvement of reading, or the involvement of an auditory component. Although these tasks are well accepted in the literature as tasks examining EF (Axelrod et al., 1994; Romine et al., 2004; Carlson, 2005), another possibility is that the decreased performance in these tasks is a basic perceptual deficit rather than a deficit in EF. However, since vast literature suggested a decreased performance in additional, more basic EF tasks, such as inhibition (Brosnan et al., 2002; Gooch et al., 2014), speed of processing (Willcutt et al., 2005; Breznitz, 2006), and attention tasks (Gooch et al., 2014), additional prospective studies should be looking at this question in depth. Our review discussed the existence of conflicting results in reading networks in all age groups, namely, the left inferior frontal gyrus showing hyperactivity in some studies while displaying hypoactivity in others.

Overall, the reviewed evidence on dyslexia indicates that reading tasks are associated with hyperactive EF-related brain networks. Such a tight link between EF and reading

TABLE 2 | Summary of studies assessing neurobiological changes during reading-related tasks along development.

Task	Children at risk (0–5)	Children with dyslexia (6–12)	Teenagers with dyslexia (13–21)	Adults with dyslexia (22+)
Narrative comprehension		Hyperactivation in the right superior frontal gyrus and right middle frontal gyrus (Horowitz-Kraus et al., 2016). Hyperactivation in the left middle/superior temporal gyri and bilateral insula, right cingulate gyrus, right superior frontal gyrus, and right parietal lobe (Rimrodt et al., 2008).		
Phonology		Hyperactivation of the left extrastriate cortex and hypoactivation of the temporal and prefrontal cortex (Backes et al., 2002). Hypoactivation of left fusiform cortex (Desroches et al., 2010). Hyperactivity in the basal ganglia and hypoactivity in the left superior temporal gyrus (Kita et al., 2013). Hypoactivation in right visual and left occipito-temporal cortex and left inferior frontal gyrus (Liu et al., 2012). Hyperactivation in the left inferior frontal gyrus (Georgiewa et al., 2002). Absence of connectivity between lateral inferior frontal cortex and the anterior occipito-temporal cortex (Olulade et al., 2015). Aberrant activation of the parieto-temporal and occipito-temporal area (Shaywitz et al., 2002). Hyperactivation in the right cerebellum (van Ermingen-Marbach et al., 2013).	Dyslexic vs. age-matched – hyperactivation in the left inferior and middle frontal gyri, caudate, and thalamus. Age- and reading-matched – hypoactivation in left parietal and fusiform (Hoeft et al., 2007). Hypoactivation of left fusiform cortex (Desroches et al., 2010). Hypoactivation in posterior areas of reading network (Brambati et al., 2006). Hypoactivation in left ventral occipito-temporal region, a left inferior parietal region, and a left inferior frontal region (Steinbrink et al., 2012).	Hypoactivation in the left medial geniculate body (Díaz et al., 2012). Asymmetry in inferior frontal gyrus (Hernandez et al., 2013). Hypoactivation in left ventral occipito-temporal region, a left inferior parietal region, and a left inferior frontal region. Hyperactivation in visual occipital regions (Wimmer et al., 2010).
Reading		Hypoactivation left visual word form area and Wernicke's area (Saralegui et al., 2014). Hyperactivation in the bilateral occipital cortex, inferior frontal regions, and inferior precentral gyrus (Seki et al., 2001).		Slow non-words – hyperactivation in the left inferior frontal gyrus and operculum (Karni et al., 2005).
Auditory stimuli	Event-related potentials mostly in right hemisphere compared to left hemisphere in controls (Lyytinen et al., 2004). No mismatch response (Zuijzen et al., 2013). Delayed P100 and P200 peaks for standard auditory stimuli (van Herten et al., 2008). Hypoactivation in bilateral occipito-temporal and left temporo-parietal regions (Raschle et al., 2012).			
Lexical-semantic priming	Absent N200 response (von Koss Torkildsen et al., 2007).			

disability highlights the importance of early assessment and intervention. Therefore, the inclusion of EF-specific neuro-behavioral testing in standard neuropsychological assessments

will open new windows on the developmental profile of dyslexia which, in turn, will provide clinicians with early identification signatures for improved diagnosis

and intervention. Such an early identification of children at risk for dyslexia will boost the implementation of interventions for EF and reading by strengthening or even altering some of the neurobiological and behavioral dysfunctions seen in dyslexia. Future research should aim to explore the conflicting results found in the literature to clarify the dyslexic profile. Specifically, it is crucial to explore specific EF networks activated alongside language networks during reading tasks to improve identification and intervention. Only after filling this needed research gap, it will be possible to design more comprehensive treatments for individuals with reading disabilities.

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Spatial Cognition in Children With Physical Disability; What Is the Impact of Restricted Independent Exploration?

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Edited by:

Silvio Ionta,
University of Lausanne, Switzerland

Reviewed by:

Yingying Yang,
Montclair State University,
United States
Nancy Rodriguez,
UMR 5506 Laboratoire
d'Informatique, de Robotique et
de Microélectronique de Montpellier
(LIRMM), France

*Correspondence:

Emily K. Farran
e.farran@surrey.ac.uk

†ORCID:

Emily K. Farran
orcid.org/0000-0001-6409-0875
Valerie Critten
orcid.org/0000-0003-2395-7887
Yannick Courbois
orcid.org/0000-0001-6625-1935
Emma Campbell
orcid.org/0000-0003-0527-648X
David Messer
orcid.org/0000-0001-9332-1265

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Emily K. Farran^{1*†}, Valerie Critten^{2†}, Yannick Courbois^{3†}, Emma Campbell^{4†} and David Messer^{2†}

¹ School of Psychology, University of Surrey, Guildford, United Kingdom, ² Faculty of Wellbeing, Education and Language Studies, The Open University, Milton Keynes, United Kingdom, ³ ULR 4072 – Psychologie: Interactions Temps Émotions Cognition, Université de Lille, Lille, France, ⁴ UCL Institute of Education, London, United Kingdom

Given the developmental inter-relationship between motor ability and spatial skills, we investigated the impact of physical disability (PD) on spatial cognition. Fifty-three children with special educational needs including PD were divided into those who were wheelchair users ($n = 34$) and those with independent locomotion ability ($n = 19$). This division additionally enabled us to determine the impact of limited independent physical exploration (i.e., required wheelchair use) on spatial competence. We compared the spatial performance of children in these two PD groups to that of typically developing (TD) children who spanned the range of non-verbal ability of the PD groups. Participants completed three spatial tasks; a mental rotation task, a spatial programming task and a desktop virtual reality (VR) navigation task. Levels of impairment of the PD groups were broadly commensurate with their overall level of non-verbal ability. The exception to this was the performance of the PD wheelchair group on the mental rotation task, which was below that expected for their level of non-verbal ability. Group differences in approach to the spatial programming task were evident in that both PD groups showed a different error pattern from the TD group. These findings suggested that for children with both learning difficulties and PD, the unique developmental impact on spatial ability of having physical disabilities, over and above the impact of any learning difficulties, is minimal.

Keywords: physical disability, learning difficulties, spatial cognition, motor, navigation, cerebral palsy

INTRODUCTION

Spatial cognition involves perceiving the location, dimension and properties of objects and their relationships to one another; it is core to everyday living, e.g., reading maps, packing a suitcase. There is a known relationship between motor competence and spatial cognition. For example, in typical infants, the emergence of independent walking predicts the development of spatial understanding about the layout of their environment (Clearfield, 2004) and locomotor experience in infancy enhances spatial cognition (Yan et al., 1998). This is supported by longitudinal evidence that the age at which walking emerges is predictive of spatial cognition at 32 months (Oudgenoeg-Paz et al., 2015). Beyond infancy, an association has been shown between motor ability and mental rotation performance in 5- to 6-year-olds (Jansen and Heil, 2010), and between motor ability and spatial navigation performance in 5- to 11-year-olds (Farran et al., 2019).

Further evidence for the relationship between the motor and spatial domains comes from individuals with physical disability (PD), including those with Cerebral Palsy. Physical Disability is a disturbance of movement and is used as an umbrella term that includes various subtypes and causal pathways. A diagnosis of Cerebral Palsy is given when the disorder of movement results from an early acquired non-progressive brain lesion (Rosenbaum et al., 2006); individuals with Cerebral Palsy also present with varied neural presentation and cognitive impairments (Ego et al., 2015; Stadskleiv et al., 2017). Stadskleiv et al. (2017) report that the majority of individuals with Cerebral Palsy in their study presented with white matter lesions. Their measure of MRI presentation was not associated with motor outcome, but was associated with level of cognitive ability.

Studies that have specifically investigated spatial cognition in children with PD have shown that this group demonstrate impaired spatial knowledge of their environment (Stanton et al., 2002; Wiedenbauer and Jansen-Osmann, 2006), and that individuals with PD present with impaired visuo-spatial perception (Stiers et al., 2002; Critten et al., 2018). In children with Cerebral Palsy, Belmonti et al. (2015) report impaired spatial memory on a table-top task and a large-scale spatial memory task. They also report an association between spatial memory and the extent of neural impairment for right-hemisphere lesions, but not for left-hemisphere lesions. They explain this with respect to evidence for right lateralization of visuospatial functions (for example, the right inferior parietal lobe; Schintu et al., 2014) and perception of self-motion (right parietal-temporal areas; Dieterich et al., 2003).

The aim of the current study was to better understand the relationship between motor and spatial ability domains by further investigating the impact of physical disability on spatial cognition while also contributing to the limited literature describing the impact of independent physical exploration on children's sense of spatial competence. With reference to physical exploration, Oudgenoeg-Paz et al. (2015) reported that scores on a self-locomotion physical exploration measure among their typically developing young participants (20 months of age) was predictive of small scale spatial cognition at 32 months [assessed with the Block Design subtest of the Wechsler Intelligence Scales for Children – Fourth Edition (WISC-IV); Wechsler, 2003]. Furthermore, investigators have shown that a child's experience of physical exploration in their local environment is related to the development of strategies required for successful navigation of space (Cornell et al., 2001). Since physical exploration is likely to be restricted in those with PD, due to their poor motor co-ordination, muscular weakness, limited sensations such as paralysis, difficulties with proprioception (perception of the body) and/or poor balance (Sit et al., 2007), comparing spatial cognition skills in children with PD and children without PD, can provide potential insight into the role of physical exploration opportunity as a causal factor in the development of spatial cognition.

In this study we focus on two groups of children with special educational needs including PD: children with PD who are wheelchair users; and those with PD who have independent locomotion. These groups differ with respect to independent

exploration because restrictions on exploration are likely to be increased for wheelchair users, especially in the early years. This is because, for wheelchair users, some activities and places are inaccessible and, although there are wheelchair users who are able to self-propel, many wheelchair users are often guided along routes by helpers who may repeat the same routes. This limits the individual's active control over their exploration. Active control was investigated by Foreman et al. (1994) who demonstrated poorer performance in a radial search task in 6-year-olds who were trained passively compared to 6-year-olds who experienced active training. For both passive and active free-choice conditions, they included a walking and a sitting (being pushed in a push chair) condition. They determined that the free-choice element, i.e., self-initiated exploration, was more important than the type of locomotion, thus emphasizing that for wheelchair users, restrictions to their autonomy of movement can negatively impact spatial cognition.

Whilst the above review demonstrates an incomplete understanding of the relationship between motor competence and spatial reasoning, there is a consistent pattern of past findings showing an association between them. To our knowledge, our study provides the first investigation of the relationship between motor impairments and small- and large-scale spatial cognition in a large group of children with PD. We included three assessments of spatial cognition. First, we used a mental rotation task, a relatively pure measure of small-scale spatial ability with no physical manipulation requirements in which participants match a rotated image to one of two mirror-imaged upright images. Uttal et al. (2013) and Newcombe (2018) refer to mental rotation as requiring intrinsic spatial coding, i.e., the within-object spatial relations that constitute the structure of the object. This spatial task activates the posterior parietal cortex (Zacks, 2008). It also taps into processes that are common to motor activity (Wohlschläger and Wohlschläger, 1998), and activates the precentral sulcus, a neural area associated with motor activity (Zacks, 2008). Particularly relevant to the current study, this brain activation from a mental rotation task is atypical in individuals with impaired motor ability (e.g., Biotteau et al., 2016; Kashuk et al., 2017). We predicted impaired mental rotation abilities in our participants with PD, relative to those with typical development and suspected that this deficit would be more evident among children with PD who were wheelchair users (for whom exploration might have been relatively limited) than among children with PD who were able to walk independently – as exploration was found to be associated with small scale spatial performance (Oudgenoeg-Paz et al., 2015).

We also included two route learning tasks; in contrast to the mental rotation task, these tasks can be classified as extrinsic spatial tasks (Uttal et al., 2013; Newcombe, 2018), i.e., requiring coding of the spatial relations between objects. The spatial programming task was a 2D route learning problem presented via a freely available Bee-Bot App. Bee-Bots are programmable robots and the Bee-Bot app was presented to children on an iPad. Participants were shown a map-like viewer-independent/allocentric perspective and asked to program the route that the Bee-Bot should take in order to arrive at a flower. This form of presentation allows the participant to view the set

of spatial relationships within the environment simultaneously, without actually navigating through the space; it provides a static view of the environment (see Uttal et al., 2006). The use of maps has been related to the development of allocentric spatial coding strategies (Uttal et al., 2006). Furthermore, the development of the ability to use allocentric coding has been associated with self-locomotion (Yan et al., 1998).

The second route learning task was presented using desktop virtual reality (VR) and thus represented a high level of physical realism. In contrast to the viewer-independent perspective presented in the spatial programming task, in this task participants viewed the environment from a viewer-centered/egocentric perspective. Participants were shown a route from A to B and asked to learn it. This perspective represents the prototypical manner in which we experience new environments; as we navigate, the relationship between ourselves and space is constantly changing, and landmarks are viewed sequentially. Desktop VR is ideally suited to this investigation because it neutralizes the demands of real-world locomotion, allowing a pure measure of spatial cognitive aspects of navigation.

The above two route learning tasks differ in their egocentric vs. allocentric representation of the environment, and the use of a map only in the spatial programming task. Landmark knowledge and route knowledge, as measured in both tasks, activate the parahippocampal gyrus (Wegman and Janzen, 2011) and the caudate nucleus respectively (Doeller et al., 2008). Allocentric coding and the development of configural knowledge, i.e., knowledge of the spatial relations between places within an environment activates the hippocampus, as part of the same interacting network (Doeller et al., 2008). Thus it is likely that the spatial programming task additionally activates the hippocampus. This is, of course, speculative without direct neural evidence.

We predicted poorer performance in the children with PD for both route learning tasks compared to a typically developing group. For the spatial programming task, this was based on the association between early locomotor experience and the development of allocentric coding (Clearfield, 2004). For the VR route learning task, this was based on previous reports of impaired spatial knowledge of large-scale environments in individuals with PD (Stanton et al., 2002; Wiedenbauer and Jansen-Osmann, 2006). Given the association between physical exploration and the development of both allocentric and egocentric spatial knowledge (Cornell et al., 2001; Oudgenoeg-Paz et al., 2015), as well as the impact of passive vs. active route learning on performance (Foreman et al., 1994), we predicted a further differentiation between children with PD who used a wheelchair vs. those who could walk independently, with the poorest performance predicted for the PD participants who used a wheelchair. This was based on the assumption that wheelchair users had relatively limited opportunity for independent exploration compared to non-wheelchair users. Due to the heterogeneity of neural damage in individuals with PD, we did not make predictions based on the neural activation of each spatial task.

We also included a memory element to the VR route learning task, in which participants were asked to recall landmarks along

the route. Whilst this had a spatial element, it could be solved using visual recognition and so we did not predict a deficit in the PD participants on this measure.

MATERIALS AND METHODS

Participants

For the mental rotation and spatial programming tasks, 51 typically developing children were recruited from mainstream schools in the United Kingdom (see Table 1). For the VR route learning task, in addition to the fifty-one TD children who completed the full battery of tasks, data was also included from TD children who had completed this task as part of a different study (Farran et al., 2019) bringing the total number of TD children to $N = 122$ for this task. The TD children ranged from 5 to 11 years, chosen to span the mental age range of the PD participants (which was lower than their chronological age, on account of their learning difficulties). This allows us to compare the performance of the PD group to what would be expected for their level of non-verbal ability, thus taking into account their learning difficulties.

Fifty-three participants with PD (all with statements of special educational needs) were recruited from two special schools in the United Kingdom. All children with PD who were invited to take part met the criteria of being able to verbally communicate (some children supported this by signing or gesturing), having the ability to use the keys on a computer keyboard (some children used a large-keys keyboard), and all had normal or corrected to normal vision. One of the authors, who was also a teacher of the children with PD, also completed the Movement Assessment Battery for Children 2 - checklist (MABC2; Henderson et al., 2007) for each participant. The MABC2 checklist is a thirty-item checklist in which the respondent rates the child's motor competence on a 4-point scale (0, 1, 2, or 3). The questions refer to motor skills such as self-care skills, classroom skills,

TABLE 1 | Participant details for the mental rotation and spatial programming tasks (mean and range).

Group	Chronological age (years; months)	BAS3 matrices ability score ¹	BPVS raw score	Movement ABC checklist. Total Motor Score
PD – wheelchair user ($N = 32$)	13;06 (5;11–18;02)	85.28 (58–139)	115.09 (50–164)	61.32 (19–87)
PD – no wheelchair use ($N = 18$)	13;10 (6;06–18;02)	98.22 (58–157)	112.28 (52–157)	19.00 (1–46)
TD ($N = 51$) ²	8;10 (5;10–11;07)	118.18 (58–163)	123.20 (78–160)	NA

¹BAS3 Matrices ability scores are derived from the first item that was assessed and are equivalent to raw scores.

²One participant in the TD group did not complete the mental rotation task. In order for the range of BAS ability scores to be similar across the groups, the three participants with the lowest BAS matrices scores in the PD groups were excluded from the sample for mental rotation and Bee-Bot analyses.

recreational skills, and ball skills. A total motor score is provided which is the sum of the thirty scores, with a higher score indicative of poorer motor performance. The MABC2-checklist correlates significantly with performance on the MABC2 test ($r = 0.38$; $p < 0.001$; Schoemaker et al., 2012) and has high construct validity (Cronbach's α : 0.94; Schoemaker et al., 2012). All participants completed the Matrices subtest of the British Ability Scale 3 (BAS3; Elliot and Smith, 2011) as a measure of non-verbal ability and the British Picture Vocabulary Scale III as a measure of verbal ability (Dunn and Dunn, 2009).

The children with PD were divided into two groups: (1) wheelchair users (used wheelchairs every day and for most of the day) and part-time wheelchair users (used wheelchairs for part of the day or the week); and (2) non-wheelchair users (although some of this group may have used wheelchairs at an earlier age) (see Table 1). A large proportion of the children with PD had received a diagnosis of Cerebral Palsy; $N = 33/34$ (97%) in the PD wheelchair group, and $N = 6/18$ (33%) in the PD no wheelchair group. Individuals with Cerebral Palsy have known deficits in visuo-spatial perception (e.g., Ego et al., 2015; Critten et al., 2018, 2019). The extent to which these deficits are independent of their motor impairment is not possible to ascertain. However, given that Cerebral Palsy is a lifelong disorder caused by cortical damage before, during or soon after birth, and the known developmental association between motor and spatial domains, it is highly likely that early disordered motor development in these participants has an impact on the development of spatial cognition (see Stanton et al., 2002), similar to that of an individual with a lifelong motor deficit without a diagnosis of cerebral palsy.

Ethical approval was obtained from the University Ethics Committee. Parental written consent and the children's verbal consent were obtained prior to testing. Children were tested individually in quiet areas or rooms in 20–30 min sessions. For each task, participants were given no help during the tasks beyond the standardized instructions. As this was part of a larger battery of tasks, children took part in approximately six sessions. The additional TD children who received the VR navigation task, the BAS3 matrices and BPVS were presented with these tasks under the same conditions (the same 17 inch laptop was used for VR navigation task, task administration was identical, and testing took part in a quiet area of the school within a 30-min testing session, as part of a larger battery of tasks).

Design and Procedure

Mental Rotation Task

This task, from Broadbent et al. (2014b), was presented on a 17 inch laptop computer. Participants viewed two mirror imaged monkeys on the top half of the screen and the test monkey on the bottom half of the screen (Figure 1) and were asked to choose which of the two monkeys on the top half of the screen matched the monkey on the bottom half of the screen. They responded by pressing one of two keys on the keyboard. A large-keys keyboard was available for children who found the laptop keys difficult to access, and two participants chose to answer by pointing, and their choices were inputted for them. There were 6 practice trials followed by 32 experimental trials. In the practice trials, the test

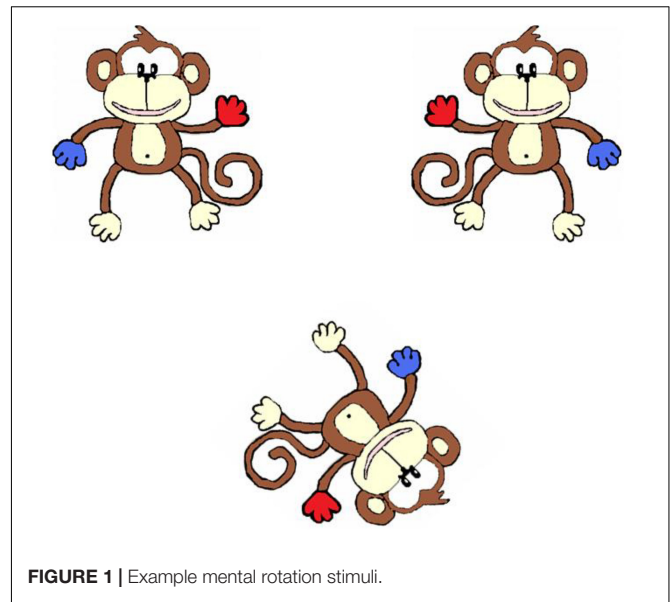


FIGURE 1 | Example mental rotation stimuli.

monkey was rotated 0° (four trials), 45° (one trial), or 90° (one trial). The practice block was repeated if participant made any errors on these trials. No feedback was given for experimental trials, but motivation language was used at the end of the task such as “Well done.” In the experimental trials, the test monkey was rotated at 0° , 45° , 90° , 135° , or 180° . Accuracy was recorded.

Spatial Programming Task

The Bee-Bot app¹ was presented on an iPad. There are twelve route planning games on the app, starting with a very simple route for the Bee-Bot to reach a flower (Figure 2). The routes gain in complexity and some routes have more than one algorithm to complete them. The first two routes were used as practice routes, and Routes 3–9 were used as experimental routes (seven routes). Participants were told that they would need to program the Bee-Bot to move it from the start along the route to the flower using the arrow keys in the corner of the screen. Participants were asked to program all moves before they started the Bee-Bot on the route by pressing the GO key. The experimental trials commenced once participants had passed the two practice trials.

Participants were told that if they made an error, they would be allowed to have another go. If participants perceived that they had made an error, motivational language was used (e.g., “Good effort”) and they were encouraged to try again. There were a maximum of five trials for each route, and if the child did not complete a route correctly within the five trials, then the task finished. The task was scored as the number of routes attempted by the children (route accuracy: max = 7). We also recorded the number and type of errors made by participants. A correct programming algorithm included two types of commands; forward displacement of the Bee-Bot and left or right 90-degree rotation of the Bee-Bot. Errors scores were coded as a proportion of errors for that command type within the route, e.g., if there were two rotation commands in a route, an

¹<https://www.tts-group.co.uk/>



FIGURE 2 | Bee-Bot app showing Routes 3 and 9. Images published with permission from TTS group (<https://www.tts-group.co.uk/>).

error of one would give a proportion of 0.5. The mean proportion error score across the number of routes attempted, for each error type, was used as the dependent variable.

VR Navigation Task

The VR navigation task was from Farran et al. (2012). Virtual environments (VEs) were created using Vizard² and presented on a 17 inch laptop computer. The VEs displayed brick-wall mazes which could be navigated using the arrow keys on the keyboard. Preceding the experimental maze, the participants watched the experimenter navigate a simple corridor that included two turns. Then they practiced navigating along the corridor. If participants had difficulty controlling their navigation, they were given another attempt.

The experimental VE displayed a brick-wall maze with 6 junctions, each leading to two paths, one correct and one incorrect. The 6 correct choices constituted two left, two right, and two straight-ahead choices. A map of the maze layout is shown in **Figure 3**. Each incorrect path choice ended in a cul-de-sac and looked like a T-junction when viewed from the preceding junction. Sixteen unique landmarks featured throughout the maze and featured equally on the left and right of the paths. Eight of the landmarks were near to junctions ('junction landmarks'). Eight of the landmarks were not near to junctions ('path landmarks'). Landmarks were selected from a range of categories (e.g., animals, tools, furniture) for their high verbal frequency (Morrison et al., 1997) and for being easy to recognize. A gray duck was shown at the end of the maze. On approaching the duck, the game ended.

Route learning task

Participants were instructed to learn a single six junction route through a maze. The experimenter showed the participant the

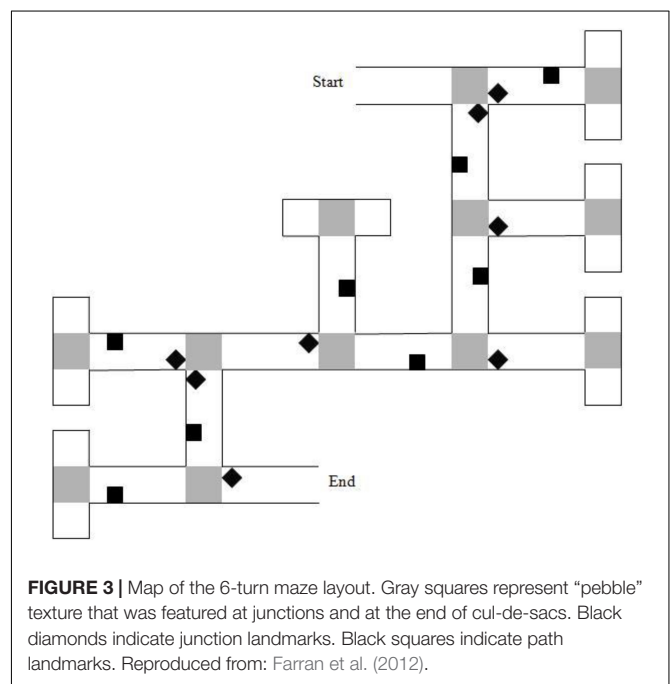


FIGURE 3 | Map of the 6-turn maze layout. Gray squares represent "pebble" texture that was featured at junctions and at the end of cul-de-sacs. Black diamonds indicate junction landmarks. Black squares indicate path landmarks. Reproduced from: Farran et al. (2012).

correct route through the maze by using the arrow keys on the keyboard to navigate and told the participant to watch, because it would be their turn to navigate next. After the experimenter demonstration, the participant attempted to walk the correct route from start to finish using the arrow keys. A large-keys keyboard was available for those children who found the laptop keys difficult to access. If the participant selected an incorrect path, they reached a cul-de-sac and could self-correct by turning around. If a participant was going backwards to the start of the maze, they were directed back to the junction where they

²<http://www.worldviz.com>

made the error. On reaching the gray duck (i.e., on completing the route) the trial terminated. Motivational language was used throughout to maintain participant concentration.

Each walk through the maze from start to finish of the route was labeled a learning trial. The criterion for having learnt the route was the successful completion of two consecutive learning trials from start to finish without error. If participants did not meet this criterion after ten learning trials, the task was stopped. The cumulative number of errors across learning trials was recorded; this was used as the dependent variable. An error was defined as a deliberate incursion down an incorrect path; if the participant corrected his/her course before reaching half-way down an incorrect path section, no error was counted.

Landmark recall task

After the participant had learnt the six junction route to criteria, they completed a landmark recall task. Participants were shown the same maze but with all landmark objects shown as red balls. The experimenter navigated, stopping at each junction to point out the red ball(s). Participants were asked to recall what object the ball had been when they were navigating the route. On providing an answer, the participant was shown a visual image of the correct answer on another computer screen as feedback, i.e., the landmark in its correct location. This feedback was given to eliminate any dependency between their answers (e.g., if the participants answered incorrectly at one location, without feedback they might not have used that landmark label again, or their incorrect answer might have negatively influenced their subsequent performance if they had recalled the landmarks in sequence). This was conducted for all 12 landmarks that were visible from the correct path. Eight of these landmarks were on the correct path, there were also four landmarks that could be viewed straight ahead before a correct turn to the left or right was executed).

To ensure that the verbal labels used by the participants in the landmark recall task could be coded accurately (e.g., a participant might use the word “light” for “streetlamp”), after the landmark recall task, participants were shown images of each of the 16 landmarks and were asked to name them. This information was then used to retrospectively facilitate the scoring of the landmark recall task.

RESULTS

Overview of Analyses

Where suitable, the data is analyzed using developmental trajectory analysis (Thomas et al., 2009). Developmental trajectory analysis does not require the individual matching of the participants and goes beyond determining differences in group means, to ascertain whether the trajectory of performance across the range of mental ages of each group differs at the onset of the trajectory (the youngest mental ages measured) or the rate of development. For developmental trajectory analysis to be meaningful, it is important that a measure of mental age (in this study, BAS3 matrices ability score) correlates with the task dependent variables. This was the case for the mental rotation

and spatial programming tasks, but not the VR navigation task. For the VR navigation task, comparison was by group means instead.

Developmental trajectory analyses were ANCOVAs with Group as the between-participant factor and BAS3 matrices ability scores as the covariate. We chose BAS3 matrices ability score (equivalent to raw score) as our measure of mental age because it is a measure non-verbal ability and thus represents ability within the same domain as the tasks of interest. BAS3 matrices ability score was rescaled so that the X-axis crossed the Y-axis at the lowest BAS matrices score (a score of 58) of the participants. That is, we subtracted 58 from all BAS matrices scores for these analyses. This does not change the analyses but is easier to interpret because the starting point for the trajectories is at zero. The ANCOVA model included interaction terms between the BAS3 matrices covariate and Group. This was used to indicate whether spatial ability developed at a different rate for each group, with respect to non-verbal ability.

The mental rotation variables were broadly normal (Kolmogorov–Smirnov, $p > 0.05$). Spatial programming and VR navigation variables were largely not normally distributed (Kolmogorov–Smirnov, $p < 0.05$). Because ANOVA is robust to violations of assumptions of normality, parametric analyses were applied (Blanca et al., 2017) with one exception, maze error. For this variable, responses were skewed toward zero, and thus non-parametric analyses were conducted. For associational analyses, parametric and non-parametric analyses were applied for normal and non-normal distributions respectively.

Mental Rotation

Developmental trajectory analysis was conducted on the proportion of correct answers with degrees of rotation (0° , 45° , 90° , 135° , 180°) as a within-participant factor and Group as a between-participant factor. This revealed the anticipated main effect of rotation (decrease in accuracy with increasing degrees of rotation), reported as a linear contrast, $F(1,94) = 18.94$, $p < 0.001$, $\eta_p^2 = 0.17$. The effect was consistent across participant groups, $F < 1$. There was no group difference in proportion correct at the lowest level of non-verbal ability (i.e., at the intercept of the

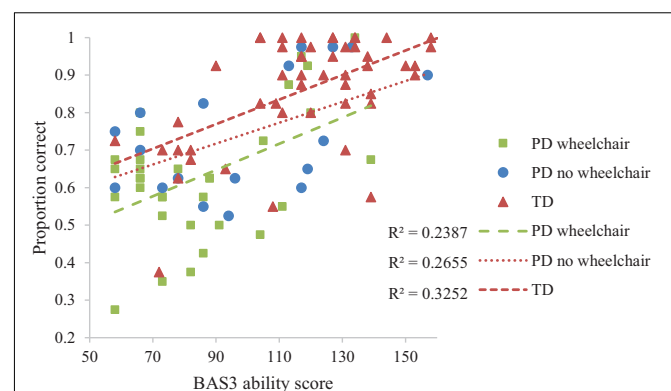


FIGURE 4 | Developmental trajectory of mental rotation performance.

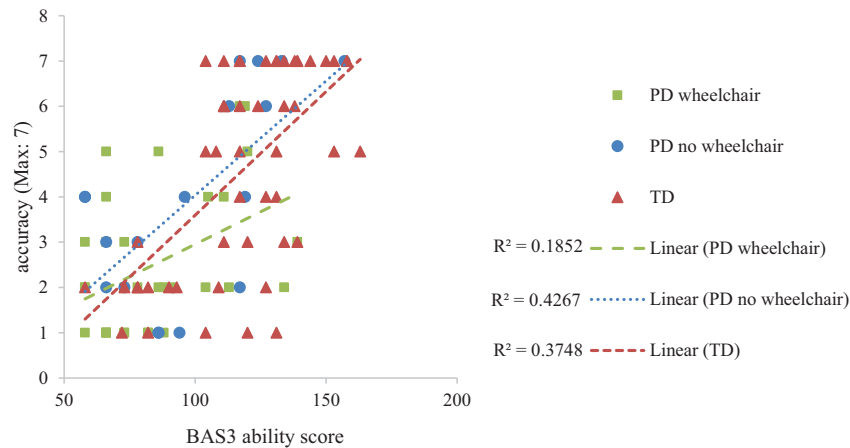


FIGURE 5 | Developmental trajectory of spatial programming accuracy.

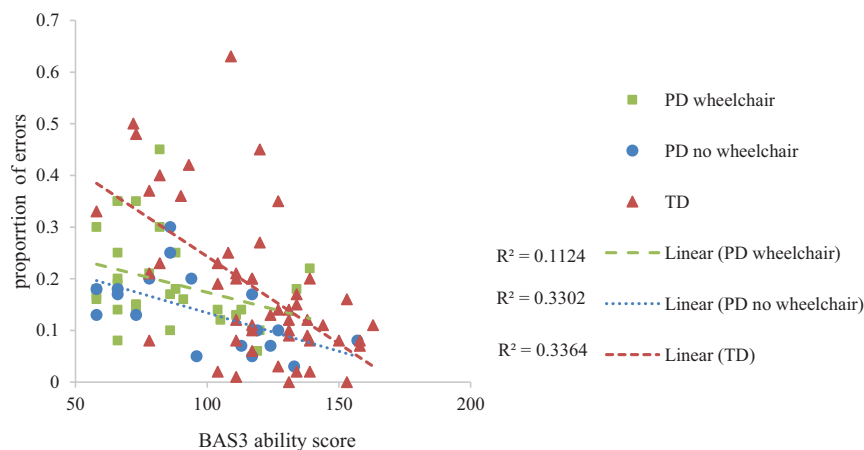


FIGURE 6 | Developmental trajectory of proportion of spatial programming errors per route attempted.

trajectories), $F(2,94) = 2.529$, $p = 0.085$, $\eta_p^2 = 0.051$). However, because this effect was marginal we had reason to explore it. This revealed a lower proportion correct at the lowest level of non-verbal ability in the PD wheelchair group compared to the TD group only ($p = 0.036$; other comparisons; $p > 0.05$; see **Figure 4**). There was no interaction between non-verbal mental age and group, which is indicative of similar rates of development across groups, $F < 1$. BAS3 matrices score (non-verbal mental age) was significantly related to proportion correct, $F(1,94) = 32.079$, $p < 0.001$, $\eta_p^2 = 0.254$. All other interactions with BAS3 matrices score were non-significant, $p > 0.05$ for all.

Spatial Programming

Spatial Programming Route Accuracy

Developmental trajectory analysis on the number of routes attempted (route accuracy, maximum = 7) with Group as a between-participant factor demonstrated no group difference at the lowest level of non-verbal ability, $F < 1$ and similar rates of development, $F(2,95) = 1.379$, $p = 0.357$, $\eta_p^2 = 0.296$

across the groups. BAS3 matrices score was significantly related to Bee-Bot route performance, $F(1,95) = 39.875$, $p < 0.001$, $\eta_p^2 = 0.296$ (**Figure 5**).

Spatial Programming Errors

Developmental trajectory analysis on proportion error scores, with a within-participant factor of Error Type (forward errors, turn errors) and Group as a between-participant factor demonstrated a group difference at the lowest level of non-verbal ability, $F(1,95) = 7.525$, $p = 0.001$, $\eta_p^2 = 0.14$ and an interaction between non-verbal ability and group, which is indicative of different rates of development, $F(2,95) = 3.20$, $p = 0.045$, $\eta_p^2 = 0.063$ across the groups. This was accounted for by significantly more errors at the intercept in the TD group compared to both of the PD groups (TD vs. PD wheelchair, $p = 0.002$; TD vs. PD no wheelchair, $p = 0.002$; PD no wheelchair vs. PD wheelchair, $p = 0.417$), and a steeper improvement with development in the TD group compared to the PD wheelchair group (TD vs. PD wheelchair, $p = 0.041$;

TABLE 2 | Participant details for the VR navigation task (mean and range).

Group	Chronological age (years; months)	BAS3 matrices ability score ¹	BPVS raw score	Movement ABC checklist Total Motor Score
PD – wheelchair user (N = 34)	13;06 (5;11–18;02)	82.91 (41–139)	115.06 (50–164)	61.03 (19–87)
PD – no wheelchair use (N = 19)	13;10 (6;06–18;02)	95.58 (48–157)	112.26 (52–157)	19.89 (1–46)
TD 5–7 years (N = 44)	6;07 (5;10–7;11)	91.27 (37–131)	99.36 (69–136)	NA
TD 8–9 years (N = 47)	8;10 (8;01–9;10)	123.64 (95–154)	125.87 (92–154)	NA
TD 10–11 years (N = 31)	10;09 (10;03–11;07)	140.55 (104–177)	141.71 (97–160)	NA

¹ BAS3 Matrices ability scores are derived from the first item that was assessed and are equivalent to raw scores.

TD vs. PD no wheelchair, $p = 0.076$; PD no wheelchair vs. PD wheelchair, $p = 0.853$). The slopes of the trajectories for each error type did not differ, $F(1,95) = 1.09$, $p = 0.30$, $\eta_p^2 = 0.011$, and this pattern was consistent across groups, $F(2,95) = 2.00$, $p = 0.14$, $\eta_p^2 = 0.040$. BAS3 matrices demonstrated a significant association with spatial programming errors, $F(1,95) = 22.39$, $p < 0.001$, $\eta_p^2 = 0.19$. **Figure 6** illustrates developmental trajectories collapsed across error type.

Navigation

A larger TD group was employed for this task, which enabled comparison with TD groups in different age ranges (**Table 2**). The PD groups had a similar level of BAS3 matrices ability score to the TD 5–7 year-olds (PD wheelchair vs. TD 5–7: $p = 0.410$; PD no wheelchair vs. TD 5–7: $p = 0.945$) and a lower level of BAS3 matrices ability score than the TD 8–9 year-olds and the TD 10–11-year-olds ($p < 0.05$ for all).

Maze Errors

Kruskal–Wallis ANOVA with a dependent variable of maze error was conducted with Group as the between participant factor. This demonstrated a main effect Group, $\chi^2(4) = 11.753$, $p = 0.019$. Mann–Whitney paired comparisons demonstrated that this was due to: (1) the PD groups making more errors than the TD 10–11 year-olds (PD wheel chair vs. TD 10–11 years, $p = 0.008$; PD no wheelchair vs. TD 10–11 years, $p = 0.015$); and (2) developmental progression across the TD groups (TD 5–7 years vs. TD 10–11 years, $p = 0.003$; TD 8–9 years vs. TD 10–11 years, $p = 0.044$) (all other comparisons, $p > 0.05$) (**Table 3**).

Landmark Recall

ANOVA of the number of junction and path landmarks that were correctly recalled was carried out, with a between-participant factor of Group and a within-participant factor of Landmark Type (path, junction). This demonstrated no difference in the number of landmarks recalled across groups, $F(4,166) = 2.093$, $p = 0.084$, $\eta_p^2 = 0.048$ (Tukey pairwise comparisons were non-significant for this marginal effect: $p > 0.05$ for all).

TABLE 3 | Cumulative number of errors made on the VR navigation task across learning trials.

Group	Median (range)
PD wheelchair	2 (0–14)
PD no wheelchair	2 (0–12)
TD 5–7 years	2 (0–7)
TD 8–9 years	1 (0–9)
TD 10–11 years	1 (0–7)

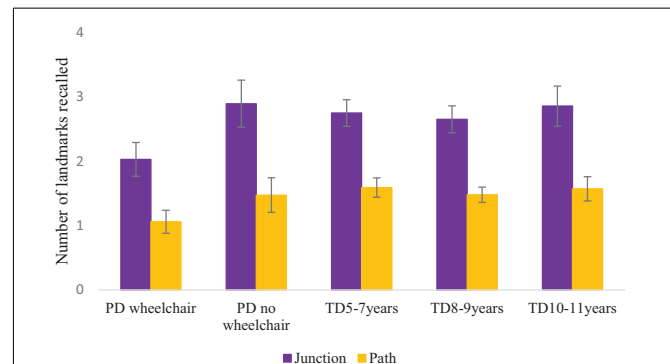


FIGURE 7 | Mean (S.E.) number of landmarks recalled per group.

TABLE 4 | Correlations between Movement ABC checklist Total Motor Score and spatial variables, for each PD group.

	Mental rotation	Spatial programming		VR navigation	
		Accuracy	Errors	Maze errors	Landmark recall
PD – wheelchair user (N = 34)	$r = -0.30$, $p = 0.09$	$+r = -0.23$, $p = 0.20$	$+r = 0.28$, $p = 0.20$	$+r = -0.30$, $p = 0.09$	$+r = -0.07$, $p = 0.69$
PD – no wheelchair use (N = 19)	$r = 0.08$, $p = 0.75$	$r = 0.03$, $p = 0.91$	$r = 0.004$, $p = 0.99$	$+r = 0.05$, $p = 0.85$	$r = -0.322$, $p = 0.18$

⁺ Indicates Spearman correlations. All remaining are Pearson correlations.

There was a main effect of landmark type due to stronger recall of junction than path landmarks, $F(1,166) = 159.463$, $p < 0.001$, $\eta_p^2 = 0.490$, which did not interact with group, $F < 1$ (**Figure 7**).

Associations Between Motor Ability and Spatial Competence

We were also interested in how performance on the M-ABC checklist correlated with each of our spatial dependent variables. M-ABC checklist data is available for the two PD groups only, and so the correlation matrix below does not include the TD group. As shown in **Table 4**, there were no significant correlations between motor score and spatial competence.

DISCUSSION

The current study had two aims. The first aim was to investigate the relationship between motor ability and spatial competence by working with participants for whom motor ability is impaired. The second aim was to investigate whether this relationship differed for those who were wheelchair users and potentially limited in opportunities for independent exploration, compared to those who could walk independently. All participants with PD had a statement of special educational needs (e.g., moderate learning difficulties, epilepsy). This was evident in their level of non-verbal ability, which was commensurate with that of TD 5- to 7-year-olds.

We predicted that the PD groups would show impaired spatial ability on all three tasks. We also predicted a differentiation in performance between the two PD groups for all three spatial tasks, with the PD wheelchair group finding the tasks harder than the PD no wheelchair group, on account of differences in their opportunities for independent exploration. We found that level of impairment in the PD groups across tasks was broadly akin to their level of non-verbal ability (note that the PD groups had poor non-verbal ability). This demonstrates that spatial ability is poor (i.e., it is not age-appropriate), but that in the context of the learning difficulties of these individuals, it does not represent a specific area of weakness. The one exception to this was performance of the PD wheelchair group on the mental rotation task, where performance was lower than expected for their level of non-verbal ability. Mental rotation taps into intrinsic spatial skills, whilst the two spatial route tasks tap into extrinsic spatial skills (Uttal et al., 2013; Newcombe, 2018). Precisely why performance on the mental rotation task and/or intrinsic spatial skills would show a specific impairment relative to the two spatial route tasks and/or extrinsic spatial skills is difficult to determine. The difference could relate to the neural activation of motor areas of the brain in the mental rotation task specifically (Zacks, 2008). However, this is a tentative explanation given the known heterogeneity in neural deficit in individuals with physical disability and learning difficulties (e.g., Stadskleiv et al., 2017).

The overall pattern of performance observed could also reflect differences in the sensitivity and specificity of the route learning tasks. The VR navigation task relied on landmark knowledge and route knowledge and thus did not draw on the more sophisticated configural knowledge. Navigational tasks that rely on configural knowledge, an ability which develops in typically developing children between the ages of 5 and 10 years (Bullens et al., 2010; Broadbent et al., 2014a), might have been more sensitive to group differences. Furthermore, neither of the route learning tasks are pure measures of spatial ability. Route knowledge tasks also draw on executive function skills (Purser et al., 2012) and we discuss below that, for the spatial programming task, the working memory and attention demands of the task might explain the pattern of errors of the two PD groups. Limitations in working memory and attention, on account of learning difficulties could thus overshadow any differences between the two PD groups in spatial competence. The pattern of spatial performance in the

PD wheelchair group is discussed further within the context of each task below.

We also predicted that performance on the landmark recall task would not be an area of deficit for the PD groups. This was the case. In fact, there were no group differences on this task, demonstrating that the mechanisms tapped into on this task (object memory) were not impacted by either physical disability or the participants' learning difficulties. Although, note that the lack of evidence in progression in the three TD groups could also suggest that this measure was not sensitive to developmental differences. Performance on each task is discussed in turn below.

Performance on the mental rotation task demonstrated a linear decrease in accuracy with increasing degrees of rotation for all groups. This pattern was expected for the TD group (e.g., Farran et al., 2001). The presence of this typical pattern for both of the PD groups suggests that the PD groups were capable of performing mental rotation and approached the task in a typical manner. Despite this, the PD groups performed at a lower level than expected for their chronological age (mean: 13 years), and at a level commensurate with their level of non-verbal mental age. A lower level of performance was observed in the PD wheelchair group, compared to the TD group from the lowest level of non-verbal ability and remained consistently low throughout the range of non-verbal abilities, as indicated by the similar rate of development to the TD group. In other words, across the range of non-verbal abilities that we examined, the PD wheelchair group was consistently and to the same degree poorer than the TD group on the mental rotation task, suggesting delayed but parallel development. In contrast, for the PD no wheelchair group, performance was on a par with the developmental trajectory of the TD group and therefore as expected for their level of non-verbal ability. Thus, any deficit in mental rotation ability in this group appears to be attributable to having learning difficulties (indexed here by non-verbal ability), rather than motor impairments. Note, these group comparisons were explored based on a marginal interaction effect and so should be considered cautiously.

The PD wheelchair group are likely to have limited experience of exploration and limited experience of actively moving through their environment. This could have a developmental cascading impact on the development of their ability to perform mental rotation. This is supported by Oudgenoeg-Paz et al. (2015) who demonstrated that exploration in TD toddlers was longitudinally predictive of their performance on a block construction task (a task which involves mental rotation; Farran et al., 2001). It is also noteworthy from the MABC-checklist scores that the PD wheelchair group had more severe motor impairment than the PD no wheelchair group. This was the case across all subsections of the checklist (Table 5), including sections A1 and A2 which included fine motor items. It is possible that this broad difference in motor competence between the PD groups, rather than or in addition to their experience of independent exploration, can explain why mental rotation was impaired in the PD wheelchair group relative to their non-verbal ability. Whilst this is not statistically supported by the correlational analyses which indicated no significant associations between motor ability and spatial competence, the relationship does show

TABLE 5 | Movement ABC checklist profile of scores for the PD groups (a higher score indicates higher severity).

	PD wheelchair user (N = 34)	PD no wheelchair use (N = 19)	Group comparison (t-test)
A1 (max: 15)	8.44 (0–15)	2.16 (0–7)	$p < 0.001$
A2 (max: 15)	9.24 (0–15)	2.32 (0–7)	$p < 0.001$
A3 (max: 15)	11.35 (5–14)	2.74 (0–8)	$p < 0.001$
B1 (max: 15)	9.41 (3–13)	1.79 (0–7)	$p < 0.001$
B2 (max: 15)	11.55 (6–15)	7.21 (1–11)	$p < 0.001$
B3(max: 15)	11.32 (5–15)	3.68 (0–7)	$p < 0.001$
Total	61.32 (19–87)	19.89 (1–46)	$p < 0.001$

A1, *Static/Predictable Movement, Self-Care Skills*; A2, *Static/Predictable Movement, Classroom Skills*; A3, *Static/Predictable Movement, PE/Recreational Skills*; B1, *Dynamic/Unpredictable Movement, Self-Care/Classroom Skills*; B2, *Dynamic/Unpredictable Movement, Ball Skills*; B3, *Dynamic/Unpredictable Movement, PE/Recreational Skills*.

a medium effect size for this group (Cohen, 1988) and the lack of significance could reflect a lack of power for these analyses. In support of a broad motor-spatial relationship, Soska et al. (2010) report a relationship between the fine motor skills required for visual-manual exploration and small-scale spatial abilities in 4.5–7.5 months-old infants. Further support is offered from evidence that mental rotation draws on mechanisms that are common to motor activity at neural and behavioral levels (Parsons et al., 1995; Zacks, 2008), supporting a direct impact of motor impairment on performance on this task for the PD wheelchair group. Further research with a larger participant group is required to determine the motor-spatial association in this context.

For both of the PD groups, performance on the navigation task was lower than the level of 10- to 11-year-old TD children, despite the age range of the PD groups spanning from 5 to 18 years. This level of navigation ability is broadly in line with the level of non-verbal ability of the two PD groups, which was similar to that of the TD 5- to 7-year-old group. The association between motor ability and performance on the VR navigation task showed a medium (albeit non-significant) effect size for the PD wheelchair group. Whilst this could be taken to suggest some impact of their motor impairment on navigation performance, the lack of group difference in navigation performance between the two PD groups suggests that the physical disabilities of the PD groups were not the limiting factor, but rather it was their learning difficulties. At first blush, this appears to contrast to previous reports of impaired navigation in people with physical disabilities (Stanton et al., 2002; Wiedenbauer and Jansen-Osmann, 2006). However, on a closer look, it simply reflects differences in the matching procedures across the studies. Stanton et al. (2002) did not measure IQ (all participants had cognitive performance in the ‘normal’ range) and matched participants by Chronological Age. Thus, their PD group performed at a lower level on a navigation task than expected for their chronological age, which is largely consistent with the current study. Furthermore, Stanton et al. (2002) also used a developmentally more sophisticated measure of navigation, which might have differentiated the groups more than the current measure of navigation. Given that a large proportion of their sample had a diagnosis which implicates poor visuospatial cognition (Cerebral Palsy or Spina Bifida), without

cognitive data it is difficult to disentangle the extent to which this contributed to their navigation performance. Wiedenbauer and Jansen-Osmann (2006) report data from children with Spina Bifida and TD controls. Their groups were matched on Chronological Age and Verbal IQ and thus the Spina Bifida group had lower non-verbal IQ than the TD control group. As such, the deficit in navigation that they report is relative to their Chronological Age and not their (lower) non-verbal ability; our data are also broadly consistent with this pattern of findings, as we observed a deficit relative to Chronological Age. One might argue that by comparing spatial performance in our sample to their level of non-verbal mental age, we are risking matching away any group differences. Whilst this is a risk, it is the most appropriate way to account for the cognitive learning difficulties of our PD samples. Furthermore, the use of developmental trajectories and error analyses in this study has enabled us to capture additional information in relation to development, individual differences and task approach.

The pattern of performance on the navigation task demonstrated that all groups had stronger recall of landmarks at junctions than landmarks on other parts of the path sections. This is in line with our predictions and suggests that all children were using a landmark strategy when learning the route, i.e., they understood that landmarks at junctions were relatively more useful for route learning than other landmarks. This strategy is consistent with the literature on the typical development of route learning (e.g., Farran et al., 2012), and appears to be robust to atypical development as it has been observed in several atypical groups including Williams syndrome (Farran et al., 2012) and children with Attention Deficit Hyperactivity Disorder (ADHD) (Farran et al., 2019). Consequently, despite having both physical disabilities and learning difficulties, these participants appeared to be able to encode landmarks effectively, and use them as a tool when navigating.

The pattern of performance of the PD groups on the spatial programming task differed from that of the TD group. For both PD groups, the number of routes attempted was in line with that expected for their level of non-verbal mental age and showed a typical rate of development. This was, however, coupled with group differences in the error patterns which suggests that the PD groups were approaching the task in a different manner to the TD group. Developmentally, at the lowest level of non-verbal ability, the TD group had higher proportion error scores than both PD groups, even though they were more successful in progressing through the routes. There are a number of reasons for this finding. A high proportion of errors could indicate a difficulty in perspective taking. For example, if the Bee-Bot is facing right, and it needs to move upwards on the iPad, the participant must determine that this requires a 90° left turn, i.e., they need to view the turn from the perspective of the Bee-Bot and not themselves. Given that perspective taking is a relatively late spatial skill to develop (Frick et al., 2014), this might have impacted the TD group more than the PD group who had more years of experience and perhaps more exposure to allocentric representations of space. The relatively late development of perspective taking (Frick et al., 2014) and processing allocentric representations (Bullens et al., 2010; Broadbent et al., 2014a) could explain why the

TD group exhibited a high number of errors at the lowest level of non-verbal ability. This contrasts to the other spatial skills measured in this study, such as mental rotation and route knowledge, which are available from at least five years in typical development (e.g., Lingwood et al., 2015). Furthermore, due to the threshold procedure employed, the TD group were exposed to a broader range of routes, and so encountered relatively more of the difficult routes (which necessarily included more changes in perspective) than the PD groups. If perceptive taking and/or allocentric coding was more problematic for those with lower non-verbal ability, this would be compounded by exposure to a larger range of routes, as observed in the TD group. The group difference at the trajectory intercept was coupled with a steeper rate of development for the TD group relative to the PD groups, which meant that the TD group caught up with the PD groups as non-verbal ability increased. This difference in the rate of development between the TD and PD groups might reflect differences in the performance limitations of each group. If the TD group are initially failing due to poor perspective taking and/or poor allocentric knowledge, their rate of development might be related to the development of these spatial skills. The PD group might have an initial advantage in these spatial skills due to their higher chronological age and level of experience with map-like representations. However, other factors might limit their progression such as juggling the spatial demands with more domain general demands such as working memory and attention, skills which might be limited in these groups due to their general learning difficulties. This might have led participants to make mistakes such as miscounting the number of paving slabs, losing where they are on the route when planning their algorithm, or forgetting the function of the buttons (e.g., understanding that the turn function programs the Bee-Bot to turn within their own square rather than moving forward one square when it turns). These kinds of limitations could be more confounding across the range of non-verbal abilities, hence the shallower rate of development in these groups. These kinds of limitations might also explain why there was no difference in performance between the two PD groups. These tentative suggestions require further research which take into account the involvement of working memory and attention processes in this task.

Whilst our findings are consistent with the conclusion that physical disability *per se* does not necessarily have a broad impact on spatial competence, it is difficult to disentangle the bi-directional developmental influence of physical disabilities and learning difficulties when both are present from birth, as in our sample. A large proportion of our sample had a diagnosis of Cerebral Palsy, which is known to present with deficits in visuospatial perception alongside motor difficulties (although note evidence for heterogeneity in visuospatial perception in Cerebral Palsy; Critten et al., 2019). We cannot rule out that any atypicalities observed in the current sample are driven by limitations in visuospatial perception that are associated with Cerebral Palsy. However, all of our PD participants had a lifelong disorder and given the known interacting developmental trajectories of the spatial and motor domains (e.g., Yan et al., 1998; Clearfield, 2004; Jansen and Heil, 2010; Oudgenoeg-Paz et al., 2015; Farran et al., 2019), further research is required to

determine any differentiated impact of a diagnosis of Cerebral Palsy, in individuals with PD and a learning disability, on spatial competence. We predict that a lifelong physical disability in *any* individual could impact the spatial domain.

To summarize, we have shown across three different spatial tasks that children with PD and learning disabilities perform lower than an age-appropriate level, but for the most part, at the level expected for their level of non-verbal mental age. Mental rotation was one exception to this finding; a skill that was particularly problematic for the children who relied on a wheelchair. We also observed unusual error patterns in both PD groups on the spatial programming task. Whilst it appears that having a physical disability did not always impact the development of spatial cognition over and above any general learning difficulties in our groups, there were indications of some minor, but potentially significant impacts of having a physical disability on spatial cognition. This highlights the importance of enabling active exploration for individuals with PD, particularly for those who are wheelchair users; evidence supports the importance of learning spatial layouts using free-choice and active exploration, over and above whether children locomote or use a wheelchair (Foreman et al., 1994).

DATA AVAILABILITY STATEMENT

The dataset presented in this study can be found in the following online repository: <https://osf.io/75skq/>.

ETHICS STATEMENT

This study was reviewed and approved by UCL Institute of Education. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

EF, VC, and DM conceived of the initial study design. VC collected the data from the Physical Disability participants. EC collected the data from Typically Developing participants. EF analyzed the data and wrote the manuscript. All the authors read, contributed to and approved the final manuscript.

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Investigating Cognitive Flexibility in Preschool Children With Autism Spectrum Disorder

Oleg Zacharov^{1*}, Rene Jürgen Huster² and Anett Kaale^{1,3}

¹ Department of Special Needs Education, University of Oslo, Oslo, Norway, ² Department of Psychology, University of Oslo, Oslo, Norway, ³ Norwegian Center of Expertise for Neurodevelopmental Disorders and Hypersomnias, Oslo University Hospital, Oslo, Norway

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*Correspondence:

Oleg Zacharov
olegjang@hotmail.com

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The current study investigated cognitive flexibility in preschool children with Autism Spectrum Disorder (ASD) and those with typical development using the Reverse Categorization (RC) task and the Dimensional Change Card Sort (DCCS) task. We further examined the relationship between non-verbal mental age (NVMA) and the performance on the two tasks. While no significant difference in performance on the RC task between the two groups was found, significantly more children in the typical developing group passed the DCCS task than children in the ASD group. NVMA was found to correlate with performance in both tasks in the typical developing group but not in the ASD group. When the children were matched on NVMA, no differences in task performance between the two groups were found. The current study found the disparity in performance in two groups on the RC and the DCCS tasks, hence illuminating the importance related to the selection of tasks when studying cognitive flexibility in preschool children with ASD. The study also cast some light on the involvement of NVMA in the performance on the RC and DCCS tasks.

Keywords: cognitive flexibility, preschool children, Autism Spectrum Disorder, typical development, non-verbal mental age

INTRODUCTION

Autism Spectrum Disorder (ASD) is characterized by deficits in social communication and interaction, and restricted and repetitive behaviors and interests (American Psychiatric Association, 2013). The term “spectrum” emphasizes that individuals with ASD exhibit wide-ranging levels of symptom severity in language and cognitive functioning. Behavioral difficulties observed in ASD, such as repetitive language and body movements, resistance to change, inflexible thinking, and problems with switching from one activity to another, are all potential indicators of impairment of cognitive flexibility (Smithson et al., 2013).

Cognitive flexibility is one of the major components of executive functioning (EF) and can be described as the ability to switch from one task to another and to quickly adjust to changes in the environment (Diamond, 2013). Cognitive flexibility may be especially important for early academic and social achievements, as it has been shown to correlate with reading comprehension (Cole et al., 2014), abstract mathematics skills (Purpura et al., 2017), and social understanding (Bock et al., 2015).

Difficulties with cognitive flexibility have been documented in persons with ASD on various performance-based EF tasks and rating scales, across different ages and levels of functioning (e.g., Faja and Dawson, 2013; Garon et al., 2017). Children with ASD as young as 3 years were shown to display difficulties on various measurements of cognitive flexibility (Garon et al., 2017). However, the results from studies comparing performance between young (3–7-year-old) typically developing (TD) children and children with ASD on measures of cognitive flexibility are somewhat inconsistent (e.g., Yerys et al., 2006; Gardiner et al., 2017).

For example, Smith et al. (2019) reported similar performance between 1½ and 3-year-old children with ASD and chronological age (CA)-matched TD children on a non-verbal eye-tracking task that assessed cognitive flexibility. However, it should be noted that the ASD group in this study had a significantly lower mental age (MA) and exhibited moderate-to-severe symptoms of ASD. Yerys et al. (2006) also reported no significant differences in performance on cognitive flexibility task between 2 and 3½ year old ASD and TD children matched for MA and CA. Finally, Gardiner et al. (2017) found that performance on task measuring cognitive flexibility, did not differ between 3½ and 7-year-old children with ASD and TD children matched on CA, IQ, and maternal education.

In contrast to the studies reporting null results, Garon et al. (2017) found that 3–6-year-old children with ASD performed worse than MA-matched TD children on the Preschool Executive Functioning Battery (PEFB) measuring, among other things, cognitive flexibility. Also, a study comparing 4–6-year-old children with ASD with CA- and non-verbal IQ (NVIQ)-matched TD children, reported that ASD group exhibited impaired performance (Valeri et al., 2019). Finally, Faja and Dawson (2013) reported that performance of 6–7½ year old children with ASD, as compared to CA and IQ matched TD group, was worse on the task measuring cognitive flexibility.

There are a number of factors that may contribute to the mixed findings within the literature. First, the majority of studies involving young children with ASD employ performance-based EF tasks that may have different levels of difficulty. For each task there is a proposed age at which children with TD are expected to exhibit performance close to or at ceiling. For example, the Reverse Categorization (RC) task (Carlson, 2005), which is purported to measure cognitive flexibility, has been used with TD children between 2 and 4 years. Another measure of cognitive flexibility, the standard Dimensional Change Card Sort (DCCS) task (Zelazo, 2006), has been used with TD children between 2 and 5 years.

Although both the RC and the DCCS are designed to measure cognitive flexibility among preschool children, they may arguably have different difficulty levels. The differences in difficulty level can be attributed to the number of sorting dimensions each task has. The RC task has one sorting dimension, namely color, while the DCCS task has two sorting dimensions, namely color and shape. This may consequentially require more cognitive resources to be deployed in order to perform the DCCS task compared to the RC task (Geurts et al., 2009). On the RC task,

TD children have been shown to exhibit near ceiling performance at 3 years of age (Carlson, 2005). On the DCCS task, the majority of 3-year-old TD children usually fail the post-switch condition of the standard version of DCCS, while many 4 and 5-year-old TD children pass it (Zelazo, 2006). This is especially important to consider when interpreting the findings of research in children with ASD. It may be the case that while some EF tasks related to cognitive flexibility may capture the children's impairment, others may not. Hence, it is important to clarify whether preschool children with ASD would elicit differential performance on the RC and DCCS tasks.

Another factor related to the mixed findings is the substantial degree of the heterogeneity of the cognitive profiles in young children with ASD. Due to the inhomogeneous cognitive profiles, it is common to match ASD and TD groups on some general ability, such as IQ or non-verbal mental age (NVMA) measured by standardized tests. Doing so, researchers are controlling for the fact that impaired performance among young children with ASD on EF tasks, including cognitive flexibility, may be a general outcome of developmental delay rather than being specific to the disorder. Although matching ASD and TD children on NVMA is commonplace in studies of EF, the contributions of NVMA on the performance on cognitive flexibility tasks remains under-researched. It has been argued that in TD children the development of cognitive flexibility is strongly associated with verbal development (Karchach and Kray, 2007). Nevertheless, previous research on young TD children has shown that NVMA, measured with the Mullen Scales of Early Learning (MSEL), positively correlated with performance on EF tasks, including cognitive flexibility (Stephens et al., 2018). Since verbal abilities in the majority of ASD population are impaired, some have suggested that cognitive flexibility may not be directly supported by verbal abilities, but instead by NVMA, since ASD individuals seems to rely more on visual rather than verbal abilities when solving EF tasks (Kunda and Goel, 2011). Indeed, in a sample of ASD individuals ranging from 5 to 19 years, Campbell et al. (2017) reported that NVMA, but not verbal mental age, play a unique role in the development of cognitive flexibility. However, the aforementioned studies by Smith et al. (2019) and Yerys et al. (2006) reported similar performance among TD children and children with ASD who had significantly lower NVMA, verbal MA and MA.

In conclusion, there seems to be a lack of research investigating the appropriateness of the cognitive flexibility tasks among preschool children with ASD. Moreover, more knowledge is needed about the relationship between NVMA and the cognitive flexibility performance among preschool children with ASD.

Rationale

The aim of the current study was to investigate and compare the performance of preschool TD children and children with ASD on two cognitive flexibility tasks, namely the RC and DCCS. Since the two tasks are assumed to have different levels of difficulty, we also examined whether the RC and the DCCS yield similar or contrasting results for a given age group. Although

both the RC and the DCCS tasks are designed for preschool children, it is expected that children would struggle more with the DCCS task since it has an extra dimension. Previous research on the appropriateness of executive functioning tasks, including cognitive flexibility, for different age groups were predominantly conducted with typically developing children (Carlson, 2005). Given that impairments in cognitive flexibility are implicated in ASD and the high interest in the topic, it is important that researchers are aware of which tasks may or may not be appropriate for the given study group. The current study would contribute to the field by illuminating the importance of choosing appropriate tasks when studying cognitive flexibility in preschool children with ASD.

In addition to investigating the appropriateness of the tasks, we examined whether there is a relationship between NVMA and the ASD and TD children's performance on the RC and DCCS tasks. In most cases, the NVMA is used as a main matching criterion in studies investigating cognitive flexibility in preschool children with ASD. However, how NVMA is implicated in cognitive flexibility performance among preschool children with ASD remains to be researched. According to previous report by Campbell et al. (2017), it is expected that in the current study the NVMA of preschool children with ASD would be associated with task performance on both tasks.

METHOD

Participants

46 preschool children were recruited for the current study, including 14 children with ASD, aged 40–68 months ($M = 56.00$, $SD = 7.96$) and 32 TD children, aged 37–59 months ($M = 48.81$, $SD = 6.95$) (Table 1). The ASD group consisted of 12 boys (85.7%) and 2 girls. The TD group consisted of 18 boys (56.3%) and 14 girls.

Children with ASD were recruited through the specialist health services, educational-psychological services and preschools in Oslo and surrounding counties, while children with TD were recruited through preschools in Oslo and surrounding counties. All children in the ASD group had received a diagnosis of ASD from the specialist health services based on a detailed clinical evaluation including interview with caretakers and multiple observations. All diagnoses were based on the International Classification of Diseases (ICD) 10 (World Health Organization (WHO), 1993). The current study did not validate the children's diagnoses. Instead, the Social Communication Questionnaire (SCQ) (Rutter et al., 2003) were filled out by the parents which informed about the ASD symptoms of the children with ASD. One participant had missing data. Otherwise, all but one child had SCQ scored

TABLE 1 | Descriptive statistics of typically developing and autism spectrum disorder groups.

	ASD ($n = 14$)	TD ($n = 32$)	t	p	Hedges' g
CA (Months)					
M(SD)	56.00 (7.96)	48.81 (6.95)	3.09	0.003	0.99
Range	40–68	37–59			
Social Communication Questionnaire—Parents					
M(SD)	18.85 (6.73)				
Range	8–29				
NVMA (Months)					
M(SD)	32.25 (8.38)	49.13 (9.22)	−5.28	$p < 0.001$	1.88
Range	23–50	29–68			
Receptive language—age (months)					
M(SD)	29.07 (13.33)	51.72 (10.27)	−6.27	$p < 0.001$	2.01
Range	13–62	27–69			
Expressive language —age (months)					
M(SD)	30.14 (17.51)	54.09 (12.69)	−5.23	$p < 0.001$	1.67
Range	14–67	26–70			
Child's spoken language					
Norwegian	8 (57.1%)	15 (46.9%)			
Norwegian + Other	2 (14.3%)	7 (21.9%)			
Missing Data	4 (28.6%)	10 (31.3%)			
Gender					
Male	12 (85.7%)	18 (56.3%)			
Female	2 (14.3%)	14 (43.8%)			
Maternal education					
Primary school	1 (7.1%)				
High school	1 (7.1%)	1 (3.1%)			
University	9 (64.3%)	21 (65.6%)			
Missing data	3 (21.4%)	10 (31.3%)			

above the cut-off for ASD (**Table 1**). In the ASD group, 57.1% of children spoke Norwegian, 14.3% spoke Norwegian and other language and 28.6% had missing data. In the TD group, 46.9% of children spoke Norwegian, 21.9% spoke Norwegian and other language and 31.3% had missing data. Children with severe motor, visual or hearing impairments were not included in the study. The study was approved by Regional Committees for Medical and Health Research Ethics and all parents provided a written informed consent.

Procedure

The current study was part of a broader longitudinal research investigating early development and learning in children with ASD and TD. All children were administered a number of tests including measures of language and cognitive abilities. Cognitive flexibility was measured with the RC task and the DCCS task. All 32 children in the TD group completed both tasks. In the ASD group, all 14 children completed the RC task, while 13 completed the DCCS task, as one ASD child was excluded from the analysis due to not satisfying the pre-requisite (discussed below) for being scored on the post-switch phase of the DCCS task. The performance on both cognitive flexibility tasks was videotaped. Testing was carried out by the first author and research assistants in a quiet room in the children's preschool or in the laboratory at the University of Oslo. Test duration for each child ranged from 2 to 4 h including breaks. Social (e.g., praise, play brakes) and edible motivators (e.g., raisins, apple bits) were provided when necessary to encourage children to complete the tasks. Demographic information was obtained via questionnaires to parents.

MEASURES

Cognitive and Language Level

The Mullen Scale of Early Learning (MSEL; Mullen, 1995) was used to estimate the children's NVMA and expressive and receptive language level. MSEL is a comprehensive test of language, cognitive and motor functioning that is individually administered to infants and children up to 68 months of age. MSEL consists of five subscales, namely Gross Motor, Fine Motor, Expressive Language, Receptive Language, and Visual Reception. The subscales can be used to calculate an Early Learning Composite Score which is analogous to the traditional IQ score. The Visual Reception and Fine Motor subscales were used to calculate the NVMA, while the Receptive and Expressive subscales were used to calculate language level.

Executive Functioning Measures

Reverse Categorization

RC is purported to measure cognitive flexibility in preschool children (Carlson, 2005). The task requires children to sort objects according to the first rule and then switch to a new sorting rule.

In the current study, children were presented with a blue and a red bucket that served as sorting containers for 18 blue and 18 red wooden blocks (**Figure 1**). There were two sorting conditions in this task, namely pre-switch and post-switch conditions. The



FIGURE 1 | Materials for the Reverse Categorization task.

pre-switch condition required children to put red blocks into a red bucket and blue block into a blue bucket. The post-switch condition required children to put red blocks into a blue bucket and blue blocks into a red bucket. Each sorting condition had 12 trials. Before the administration of the task, children were provided with minimal verbal instructions ("red in red bucket and blue in blue bucket") and four demonstration blocks (2 red and 2 blue) were sorted by the experimenter. After the demonstration phase, to ensure that the task was understood, children sorted four practice blocks with a rule repeated before every trial. Upon completing the practice session, 12 pre-switch blocks (6 red and 6 blue) were then handed to children one by one in a random order with the rule repeated before every third trial. After completing 12 pre-switch trials, children were informed about the second rule ("red in blue bucket and blue in red bucket"). The experimenter sorted four demonstration blocks while repeating the new rule. After the demonstration phase, no practice session was administered and children were handed 12 post-switch blocks (6 red and 6 blue) one by one in a random order with the rule repeated before every third trial.

Video recordings of children's performance were coded using VLC media player (VideoLan, 2006) and scored by the first author. For each trial the score of 1 was assigned if the child sorted the block according to the rule. The score of 0 was assigned if the child either (1) placed the block into the wrong bucket, or (2) placed the block into the wrong bucket but then took the block out and placed it in the correct bucket. In accordance with established procedures of the task, placing the block in either of the buckets meant the end of the trial, the following

actions were disregarded and hence the score of 0 was assigned for the trial in which the child placed a block in a wrong bucket. In order to be scored on post-switch phase of the RC task, children had to correctly sort 10 out of 12 pre-switch trials (Carlson, 2005). The dependent variable was the total number of correctly sorted post-switch trials. This total score was analyzed both categorically, as passing or failing the task, and subjected to Spearman's correlation analyses. In order to pass the task, children were required to sort correctly minimum 10 out of 12 post-switch blocks (Carlson, 2005). For the assessment of inter-rater reliability, a trained research assistant who was blind to the participants' group double-coded 43.48% ($n = 20$) of the RC task recordings. Scoring of the number of sorted pre- and post-switch trials was found to have high reliability ($\kappa = 0.879$, 95% CI [0.722–1], $p < 0.001$).

Dimensional Change Card Sort

DCCS is purported to measure cognitive flexibility (Zelazo, 2006). There are two versions of the DCCS task, namely the standard version, which was used in the current study and the border version which is suitable for older children due to increased complexity. The standard version requires participants to sort a number of cards according to first dimension (e.g., shape), and then according to the second dimension (e.g., color).

In the current study, children were presented with two gray plastic opaque containers. Each container had a slot at the top and a target card (dimensions) depicting either a red fish or a blue cow attached at the back (Figure 2). In total, there were 22 laminated cards depicting blue/red fish and blue/red cow on a white background. Specifically, there were 7 cards depicting a blue fish, 4 depicting a red fish, 4 depicting blue a cow, and 7 depicting a red cow. The task has two sorting conditions: a pre-switch condition, where the cards are sorted according to the first dimension and a post-switch condition, where the cards are sorted according to the second dimension. Each sorting condition had 5 trials. Prior to the experiment, minimal verbal instructions ("red animals go here and blue animals go here" or "fish goes here and cow goes here") were given and 4 demonstration cards were sorted by the experimenter. To ensure that the task was understood, 4 practice cards were sorted by children with a rule repeated before every trial and each card verbalized by the experimenter. Upon completing the practice session, children were required to sort 5 pre-switch cards. For the "color" dimension, the cards were handed in the following order one by one: blue fish, red cow, red fish, blue fish, and red cow. For the "shape" dimension, the order was as follows: red cow, blue fish, blue cow, red cow, and blue fish. Once all 5 pre-switch cards were sorted, the experimenter informed about the switch ("now we switch"). Children were provided with a new rule and watched the experimenter sort 4 demonstration cards. No practice session was administered for the post-switch condition. Children were handed 5 post-switch cards, one at the time, with the rule repeated before every trial and each card verbalized by the experimenter.

Video recordings of children's performance were coded using VLC media player and scored by the first author. For each trial the score of 1 was assigned if the child sorted the card according to the rule. The score of 0 was assigned if the child placed the



FIGURE 2 | Materials for the Dimensional Change Card Sort task.

card into the wrong container. In order to be scored on post-switch phase of the DCCS task, children had to correctly sort 4 out of 5 pre-switch trials (Zelazo, 2006). Children who did not satisfy this criterion were excluded from the analysis. The dependent variable was the total number of correctly sorted post-switch trials. This total score was analyzed both categorically, as passing or failing, and subjected to Spearman's correlation analyses (Zelazo, 2006). In order to pass the task, children were required to sort correctly minimum 4 out of 5 post-switch cards. For calculation of inter-rater reliability, 43.48% ($n = 20$) of the DCCS task recordings were double-coded by the trained research assistant who was blind to participants' group. Scoring of the number of sorted pre- and post-switch trials was found to have high reliability ($\kappa = 1$, $p < 0.001$).

Statistical Analysis

Statistical Package for the Social Sciences (SPSS) 27 was used to analyze the data. Descriptive data on characteristics (e.g., age, language level, gender) of the ASD and TD groups is presented as means, standard deviations and ranges or frequency and percentages. Independent sample *t*-tests were used to investigate potential group differences in these characteristics. A chi-square test was run separately for the RC and DCCS tasks to investigate whether the number of children passing the task was significantly different between ASD and TD groups and whether

TABLE 2 | Descriptive statistics of typically developing and autism spectrum disorder groups matched on non-verbal mental age.

	ASD (<i>n</i> = 9)	TD (<i>n</i> = 9)	<i>t</i>	<i>p</i>	Cohen's <i>d</i>
CA (Months)					
M(SD)	57.44 (9.06)	44.33 (6.57)	3.51	0.003	1.66
Range	40–68	37–56			
NVMA (Months)					
M(SD)	39.66 (6.47)	41.33 (5.67)	−0.581	0.570	0.27
Range	30–50	31–49			
Receptive language—age equivalent					
M(SD)	36 (11.42)	41.66 (9.04)	−1.16	0.260	0.55
Range	27–62	27–53			
Expressive language—age (months)					
M(SD)	36.88 (18.61)	43.55 (11.08)	−0.923	0.370	0.43
Range	17–67	26–60			

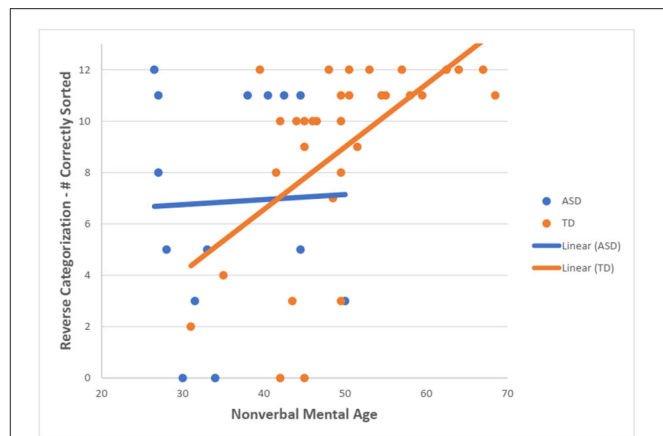
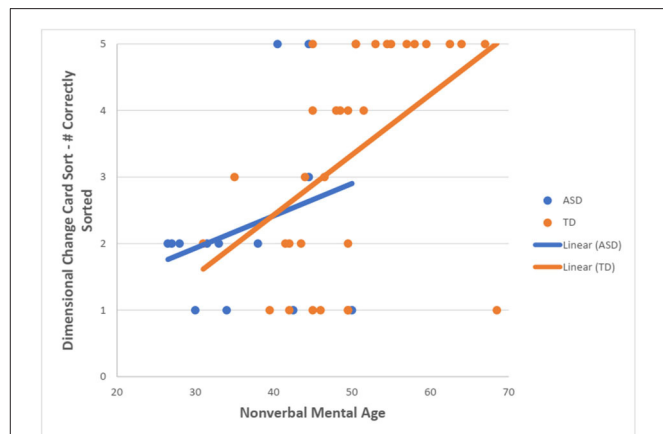
TABLE 3 | Performance of typically developing and autism spectrum disorder groups on the reverse categorization and the dimensional change card sort tasks.

Task	Group	
	ASD (<i>n</i> = 14)	TD (<i>n</i> = 32)
Reverse categorization		
# and (%) Pass	6 (42.9%)	21 (65.6%)
NVMA—M (SD)	36.50 (7.85)	52.85 (8.17)
# and (%) Fail	8 (57.1%)	11 (34.4%)
NVMA—M (SD)	34.75 (8.19)	43.81 (6.31)
Dimensional change card sort		
# and (%) Pass	2 (15.4%)	17 (53.1%)
NVMA—M (SD)	42.50 (2.82)	54.06 (6.50)
# and (%) Fail	11 (84.6%)	15 (46.9%)
NVMA—M (SD)	35 (7.79)	44.86 (8.34)

Groups are not matched on non-verbal mental age.

the children's performance on the RC and the DCCS were similar or contrasting.

Spearman's correlation analyses were preformed to determine the relationship between NVMA and the total number of correctly sorted post-switch trials on the RC and the DCCS tasks, respectively, for both groups. In these analyses the scores from the RC task and the DCCS task were used as discrete data. Finally, the study participants were matched on NVMA resulting in 9 participants both in the ASD and the TD group (Table 2). A frequency analysis was then run separately for the RC and DCCS tasks to identify number of children passing/failing the tasks in each of the matched groups. Last, Spearman's correlation analyses were run to investigate the relationship between NVMA and the total number of correctly sorted post-switch trials on RC and DCCS for the matched groups.

**FIGURE 3 |** Scatterplot showing the relationship between NVMA and a number of correctly sorted post-switch RC trials for both groups.**FIGURE 4 |** Scatterplot showing the relationship between NVMA and a number of correctly sorted post-switch DCCS trials for both groups.

RESULTS

Group Differences on the RC and DCCS Tasks

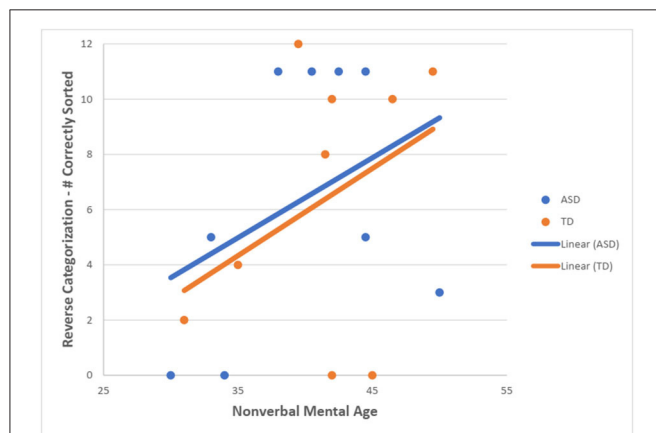
For the TD group, 21 children (65.6%) passed the RC task while 11 (34.4%) did not (Table 3). For the ASD group, 6 children (42.9%) passed the same task and 8 (57.1%) did not. A chi-square analysis revealed no significant differences in performance on the RC task between the groups ($X^2(1, n = 46) = 2.08, p = 0.149$).

For the TD group, 17 children (53.1%) passed the DCCS task while 15 (46.9%) did not (Table 3). For the ASD group, only 2 children (15.4%) passed the DCCS task and 11 (84.6%) did not. A chi-square analysis revealed that TD children were significantly more likely than children with ASD to pass the DCCS task ($X^2(1, n = 45) = 5.39, p < 0.05$). For the effect size measure, a Phi Coefficient was run revealing a moderate effect size ($\phi = 0.346$).

TABLE 4 | Performance of typically developing and autism spectrum disorder groups on the reverse categorization and the dimensional change card sort tasks.

Task	Group	
	ASD (<i>n</i> = 9)	TD (<i>n</i> = 9)
Reverse categorization		
# and (%) Pass	4 (44.4%)	4 (44.4%)
NVMA—M (SD)	41.37 (2.78)	44.37 (4.47)
# and (%) Fail	5 (55.6%)	5 (55.6%)
NVMA—M (SD)	38.30 (8.52)	38.90 (5.72)
Dimensional change card sort		
# and (%) Pass	2 (22.2%)	2 (22.2%)
NVMA—M (SD)	42.50 (2.82)	47.25 (3.18)
# and (%) Fail	7 (77.8%)	7 (77.8%)
NVMA—M (SD)	38.85 (7.14)	39.64 (5.12)

Groups are matched on non-verbal mental age.

**FIGURE 5 |** Scatterplot showing the relationship between NVMA and a number of correctly sorted post-switch RC trials for both groups matched on NVMA.

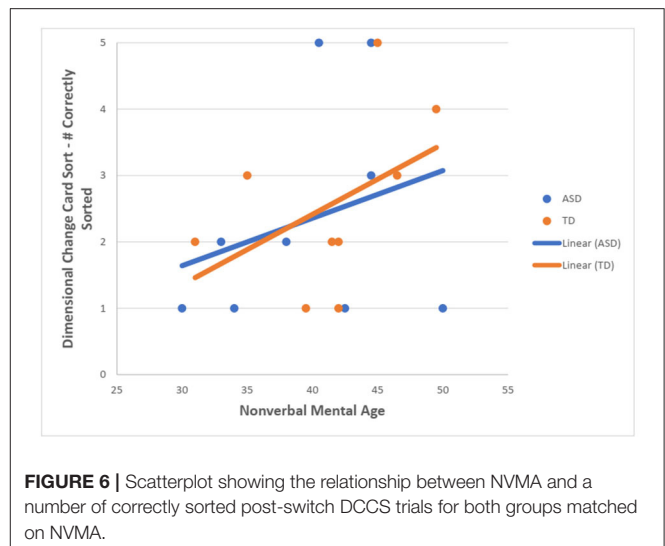
Relation Between NVMA and Performance on the RC and DCCS Tasks

For the TD group, there was a statistically significant moderate, positive correlation between NVMA and the total number of correctly sorted post-switch trials on the RC task ($r_s = 0.635$, $N = 32$, $p < 0.001$). For the ASD group, no statistically significant correlation was found ($r_s = -0.127$, $N = 14$, $p = 0.665$) (Figure 3).

For the TD group, there was a statistically significant moderate, positive correlation between NVMA and the total number of correctly sorted post-switch trials on the DCCS task ($r_s = 0.592$, $N = 32$, $p < 0.001$). For the ASD group, no statistically significant correlation was found ($r_s = 0.107$, $N = 13$, $p = 0.727$) (Figure 4).

NVMA—Matched Groups

The matched groups had equal number of children passing (TD group = 44.4%; ASD group = 44.4%) and failing (TD group =

**FIGURE 6 |** Scatterplot showing the relationship between NVMA and a number of correctly sorted post-switch DCCS trials for both groups matched on NVMA.

55.6%; ASD group = 55.6%) the RC task (Table 4). Similarly, both groups had equal number of children passing (TD group = 22.2%; ASD group = 22.2%) and failing (TD group = 77.8%; ASD group = 77.8%) the DCCS task. Finally, as illustrated in Figures 5, 6 neither of the matched groups showed significant correlations between NVMA and RC scores (TD group: $r_s = 0.194$, $p = 0.617$; ASD group: $r_s = 0.344$, $p = 0.365$) or NVMA and DCCS scores (TD group: $r_s = 0.511$, $p = 0.160$; ASD group: $r_s = 0.251$, $p = 0.514$).

DISCUSSION

The current study investigated whether the performance of preschool children with ASD, as compared to children with TD, was significantly different on two measures of cognitive flexibility, namely the RC task and DCCS task, which have been shown to have different difficulty levels. As for the RC task, which is presumably easier than DCCS task, no statistically significant differences in the number of children passing the task between the two groups were found. In both groups approximately half the children passed the RC task. In contrast, the performance of the ASD group, as compared to the TD group, was significantly lower on the DCCS task. Approximately half the children in the TD group satisfied the passing criterion, while only 15% of the children in the ASD did, despite the fact that the TD children were younger than the ASD group. The results from current study add to the findings of Faja and Dawson (2013) where older children with ASD were shown to exhibit impaired performance on the DCCS task.

Although both the RC and DCCS tasks are developed to measure cognitive flexibility in preschool children (Carlson, 2005) they may reveal different results, not only in children with TD but, as shown in this study, also in children with ASD. We found that slightly more TD children passed the RC task (65.6%) compared to the DCCS (53.1%) task. A similar, but stronger pattern was true for the children with

ASD, where almost half passed the RC task (42.9%) and only a few passed DCCS task (15.4%). These results suggest that both tasks, while being designed for preschoolers, challenge children's cognitive flexibility differently. It could be that having two sorting dimension (i.e., color and shape) the DCCS task is more challenging than the RC task because it would presumably require more attentional and/or working memory resources (Geurts et al., 2009). Hence, it may be important to consider the choice of task when studying cognitive flexibility in preschool children, and especially children with ASD. Some tasks may not be suitable for the specific age group or sensitive enough to capture the impairment leading to conclusions that are different from studies using more appropriate measures.

The current study also investigated the relationship between the children's NVMA and their scores on the RC and the DCCS tasks. For the RC task, TD group demonstrated that higher NVMA was correlated with a higher number of correctly sorted post-switch trials. No correlation for the ASD group was found. For the DCCS task, TD children with higher NVMA were more likely to pass the post-switch phase. As for the ASD group, no correlation between NVMA and DCCS scores was found. Previous research demonstrated that most of the TD preschoolers who pass the post-switch condition of DCCS were above 48 months of age (Zelazo, 2006). In the current study, CA and NVMA were shown to strongly correlate in the TD group and the mean NVMA of TD children passing the task was above 48 months while the mean NVMA of TD children who failed the task was below the age of 4. Interestingly, two children in the ASD group who passed the task had mean NVMA of 42.50, which is lower than the level at which children are expected to master the task. Furthermore, the majority of children with ASD who failed the DCCS task, despite having higher CA than TD group, had lower NVMA than those who failed the task in the TD group. Hence, it may be safe to assume that NVMA is implicated in the performance of DCCS. Similarly, NVMA may also be implicated in the RC task as it requires lower levels of NVMA to be able to satisfy the passing criterion, which would also support the notion that RC task is less demanding than DCCS task. Contrary to the Campbell et al. (2017), the findings in the current study cannot suggest that NVMA played a unique role in the cognitive flexibility performance in participants with ASD. While it is true for the TD children, the NVMA had no relationship with the performance on the cognitive flexibility tasks in the ASD group. However, this could be attributed to the small number of participants in the ASD group. With more children in the ASD group, the pattern would maybe be clearer. The current findings could also comment on the study by Yerys et al. (2006) who reported similar performance among TD children and children with ASD who had significantly lower NVMA, verbal MA and MA. As shown in the current study, both groups exhibited similar performance on the RC task while being significantly different in the NVMA. It could be that despite having lower NVMA than the TD group, the ASD group had sufficient NVMA for passing the tasks used in the Yerys et al. (2006) study.

Finally, the current study also matched the groups on NVMA. Although the matched ASD group had significantly higher

CA, the performance on RC and DCCS tasks between the two groups were found to be identical. Approximately half the children in each group passed the RC task, while 22.2 percent in both groups passed the DCCS. This further illustrate the possible contributions of NVMA on the performance on measures of cognitive flexibility and that the RC and the DCCS tasks might both be valuable measures of cognitive flexibility in young children with ASD, but that caution is needed in selecting what measure to use. It is important to note, however, that in controlling for NVMA in the matched groups the majority of good-performing TD and a number of bad-performing children with ASD were removed. This would seem to bias the performance measures, despite the fact that children with ASD had a higher CA from the beginning. This would potentially explain the correlation differences seen between NVMA unmatched and NVMA matched groups.

One of the weaknesses of the current study is the small sample size of the ASD group. Hence, caution is needed in interpreting the results. Also due to the small number of children with ASD, and the skewed gender ratio in this population, very few girls were included. Thus, the current study did not investigate potential gender differences in cognitive flexibility although gender differences related to cognitive flexibility have been previously reported (Memari et al., 2013). It is recommended that in future studies investigating cognitive flexibility in preschool children with ASD, sex differences are considered. Despite weaknesses, the current study illuminates some potential problems related to the selection of tasks when studying cognitive flexibility in preschool children with ASD. In future studies, it is recommended to use a broader set of tasks capturing the fine-tuned development of cognitive flexibility during the preschool years. In addition, the study casts some light on the involvement of NVMA in the performance on the RC and DCCS tasks. Given the vast selection of EF tasks, future studies with a larger sample that are matched both on CA and NVMA are needed to investigate tasks that measure different components of EF. There are some clinical implications of the study. First, professionals who want to measure cognitive flexibility in young children with ASD should be critical to what task they use. Cognitive flexibility tasks with less dimensions, such as the RC, might be the first choice for young children with ASD, as tasks with more dimensions, such as DCCS might be too advanced for many. Second, not only the children's chronological age, but also their non-verbal mental age should guide the selection of tasks. Last, as the findings suggest that young children with ASD have more difficulties with cognitive flexibility compared to TD peers, whether these difficulties are related to ASD or more general developmental delay, it is important to adapt the early education setting to accommodate these difficulties.

DATA AVAILABILITY STATEMENT

The data analyzed in this study is subject to the following licenses/restrictions: Not available due to restrictions related to

ethical regulations. Requests to access these datasets should be directed to Anett Kaale, anett.kaale@isp.uio.no.

ETHICS STATEMENT

The current study was reviewed and approved by Regional Committees for Medical and Health Research Ethics. Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

The data collection in the current study was conducted by OZ. All authors contributed in writing the manuscript and took part in the design and analyses of this study.

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Dorsal and Ventral Stream Function in Children With Developmental Coordination Disorder

Serena Micheletti^{1*†}, Fleur Corbett^{2†}, Janette Atkinson², Oliver Braddick³, Paola Mattei¹, Jessica Galli^{1,4}, Stefano Calza⁵ and Elisa Fazzi^{1,4}

¹ Unit of Child Neurology and Psychiatry, ASST Spedali Civili of Brescia, Brescia, Italy, ² Faculty of Brain Sciences, University College London, London, United Kingdom, ³ Department of Experimental Psychology, University of Oxford, Oxford, United Kingdom, ⁴ Department of Clinical and Experimental Sciences, University of Brescia, Brescia, Italy, ⁵ Unit of Biostatistics and Bioinformatics, Department of Molecular and Translational Medicine, University of Brescia, Brescia, Italy

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Edited by:

Jane Elizabeth Aspell,
Anglia Ruskin University,
United Kingdom

Reviewed by:

Stefania Maria Bova,
Neurologia Pediatrica - Ospedale
Vittore Buzzi, Italy
Benjamin Thompson,
University of Waterloo, Canada

*Correspondence:

Serena Micheletti
serena.micheletti@unibs.it

[†] These authors have contributed
equally to this work and share first
authorship

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Dorsal stream cortical networks underpin a cluster of visuomotor, visuospatial, and visual attention functions. Sensitivity to global coherence of motion and static form is considered a signature of visual cortical processing in the dorsal stream (motion) relative to the ventral stream (form). Poorer sensitivity to global motion compared to global static form has been found across a diverse range of neurodevelopmental disorders, suggesting a “dorsal stream vulnerability.” However, previous studies of global coherence sensitivity in Developmental Coordination Disorder (DCD) have shown conflicting findings. We examined two groups totalling 102 children with DCD (age 5–12 years), using the “Ball in the Grass” psychophysical test to compare sensitivity to global motion and global static form. Motor impairment was measured using the Movement-ABC (M-ABC). Global coherence sensitivity was compared with a typically developing control group ($N = 69$) in the same age range. Children with DCD showed impaired sensitivity to global motion ($p = 0.002$), but not global form ($p = 0.695$), compared to controls. Within the DCD group, motor impairment showed a significant linear relationship with global form sensitivity ($p < 0.001$). There was also a significant quadratic relationship between motor impairment and global motion sensitivity ($p = 0.046$), where poorer global motion sensitivity was only apparent with greater motor impairment. We suggest that two distinct visually related components, associated with global form and global motion sensitivity, contribute to DCD differentially over the range of severity of the disorder. Possible neural circuitry underlying these relationships is discussed.

Keywords: developmental coordination disorder, motion sensitivity, form sensitivity, dorsal stream, ventral stream

INTRODUCTION

Measures of coherence sensitivity to global visual form and global visual motion have been proposed as indicators of functioning in the ventral and dorsal cortical streams, respectively (Atkinson et al., 1997; Gunn et al., 2002; Braddick et al., 2003; Atkinson, 2017a). In typically developing infants and children these measures have been used to define the developmental

trajectory of sensitivity to global visual motion and global visual form (Gunn et al., 2002; Atkinson and Braddick, 2005; Braddick et al., 2016). The maturation of global visual motion sensitivity is both delayed and more variable than that found for the maturation of global visual form sensitivity across a diverse range of neurodevelopmental disorders (Atkinson, 2017b), such as Developmental Dyslexia (e.g., Hansen et al., 2001; Conlon et al., 2009), Autism Spectrum Disorders (e.g., Spencer et al., 2000; Pellicano et al., 2005; Milne et al., 2006), Williams syndrome (e.g., Atkinson et al., 1997, 2006; Atkinson, in press), Fragile X syndrome (e.g., Kogan et al., 2004), and also prematurity (e.g., Guzzetta et al., 2009; Taylor et al., 2009), and hemiplegia (e.g., Gunn et al., 2002). These findings of greater deficits in sensitivity to global motion compared to global static form have led to the concept of “dorsal stream vulnerability,” a cluster of deficits in not only global visual motion perception but also visuomotor actions and visual attention (Braddick et al., 2003; Atkinson and Braddick, 2011; Atkinson, 2017b). The present study examined the concept of dorsal stream vulnerability in children with Developmental Coordination Disorder (DCD).

DCD is a neurodevelopmental disorder characterised by a primary motor impairment affecting up to 5–6% of children (Blank et al., 2019). It is marked by clumsiness, lack of coordination, and poor balance, which negatively and persistently affect activities of daily living (American Psychiatric Association (APA), 2013). The aetiology of DCD is currently thought to be multifactorial as no single cause has been identified; both genetic and environmental influences have been implicated (Gomez and Sirigu, 2015). The prevalence of DCD is higher in males than females, with estimates of two to three males for every female diagnosed (Lingam et al., 2009; Faebo Larsen et al., 2013). For a diagnosis of DCD to be made, symptoms must be present in early childhood and motor impairments must be in excess of those associated with any intellectual disability (American Psychiatric Association (APA), 2013). The Movement-ABC (M-ABC; Henderson and Sugden, 1992; Henderson et al., 2007) is the most commonly used test battery for the assessment of motor impairment in DCD (Blank et al., 2019).

Factors that may contribute to DCD symptomatology have been examined in meta-analyses conducted by Wilson and McKenzie (1998) and Wilson et al. (2013). Wilson et al. (2013) reported large mean effect sizes ($d_w > 1.2$) for visual perceptual tasks and complex visuospatial tasks involving motor components. Visuomotor coordination is one of the primary functions of the dorsal stream, alongside visuospatial skills and the control of attention (Atkinson, 2000, 2017b; Kravitz et al., 2011). Dorsal stream processing of global visual motion provides cues necessary for detecting direction of heading and for accurate locomotion, postural control and gross motor skills (Burr et al., 1998, 2001; Gepner and Mestre, 2002). Indeed, two recent studies have shown that global motion coherence sensitivity is related to visuomotor performance in children (Braddick et al., 2016; Chakraborty et al., 2017). Given that visuomotor coordination is one of the primary functions of the dorsal stream, it might be expected that children with DCD would share with other neurodevelopmental disorders poor sensitivity to motion coherence relative to form

coherence. However, previous studies that have specifically explored global form and global motion sensitivity in DCD, using a matched groups approach, have given inconclusive results (Johnston et al., 2017).

O'Brien et al. (2002) reported that whilst global translational motion sensitivity was slightly better for eight 7–11 year olds with developmental dyspraxia compared to 50 controls, global form sensitivity was poorer. Sigmundsson et al. (2003) reported that 13 “clumsy” 10 year olds demonstrated both poorer global form and global translational motion sensitivity than typical controls. However, Sigmundsson et al. (2003) did not explicitly test the interaction between group and coherence sensitivity to form and motion to examine whether motion sensitivity was significantly lower than form sensitivity, relative to controls. The relevance of the results of O'Brien et al. (2002) and Sigmundsson et al. (2003) to DCD is difficult to assess as participants had not received a formal diagnosis of DCD and in the Sigmundsson et al. (2003) study, were allocated to groups by their performance on the M-ABC alone.

In a comparison of eleven 6–12 year olds with a diagnosis of DCD and controls, Wilmut and Wann (2008) reported that global concentric form and rotational motion sensitivity did not differ between groups. Purcell et al. (2012) compared radial (looming) motion sensitivity in eleven 6–11 year olds with DCD to controls, reporting that children with DCD showed a marked impairment in sensitivity to radial motion speed. This impairment was particularly evident when the moving object was not fixated within central vision, but no comparative measurement of form sensitivity was obtained.

These previous studies examining global form and motion sensitivity in DCD have involved only small numbers of participants ($N = 8–13$) with differing criteria for inclusion and exclusion as cases of DCD. Furthermore, in many of these studies different stimuli and tasks were used for comparing global form and motion sensitivity (Johnston et al., 2017). Sigmundsson and Haga (2010) recommend all DSM criteria to be checked, alongside the assessment of motor competency, in order for a diagnosis of DCD to be made.

The present study aimed to examine whether children with DCD show dorsal stream vulnerability as indexed by a selective impairment in global motion coherence sensitivity, compared to global form coherence sensitivity. A much larger sample ($N > 100$) of children with DCD was tested than in earlier reports, recruited in two studies, one in London, United Kingdom and one in Brescia, Italy. Children with DCD in both studies met defined DSM-5 criteria for DCD, which were validated by their scores on the M-ABC. Global form and motion sensitivity were assessed with the “Ball in the Grass” test, which is suitable for children as young as 4 years and for which extensive normative data exist (Atkinson and Braddick, 2005; Braddick et al., 2016). The size and age range (5–12 years) of the DCD group made it possible to examine (a) whether there is any association of global form and motion coherence sensitivity with individuals' level of motor deficit as reflected in M-ABC scores; (b) whether this relationship is age-dependent; (c) whether the relationship is influenced by overall cognitive ability.

MATERIALS AND METHODS

Ethical Approval

Full ethical approval was granted for the London study by the UCL ethics committee (2807/002) and for the Brescia study by the ethics committee of Brescia (NP 3513). Before advertising the research to individuals with DCD in the United Kingdom, the research was granted additional approval from the Dyspraxia Foundation Ethics Committee.

Participants

Children With Developmental Coordination Disorder (Developmental Coordination Disorder Group)

Children with a diagnosis of DCD were recruited in London (London Group $N = 17$) and Brescia, Italy (Brescia Group $N = 85$). The enrolled children had to meet the following criteria: English/Italian native speakers, aged 5:0–12:11 years old, good binocular visual acuity (≥ 0.8) to easily detect the stimuli, with performance on the M-ABC lower or equal to the 15th percentile. All children had either normal vision or no history of visual problems beyond corrected refractive errors and had no prior experience in visual psychophysics testing. Participation was voluntary, and children with DCD and their caregivers were reimbursed for their travel expenses in the London study. Details of the groups are presented in **Table 1**.

It should be noted that a lower performance on the total impairment score than on individual subsection scores, as seen here, is a common feature of M-ABC data sets (e.g., Ashkenazi et al., 2013; Romeo et al., 2018; Ricci et al., 2021) and of the published norms, presumably reflecting the incomplete correlation of the tests in each subsection.

In London, children with DCD were recruited through advertisements placed with the Dyspraxia Foundation. All had received a diagnosis of DCD, by consultant paediatricians or occupational therapists and met DSM-5 criteria (American Psychiatric Association (APA), 2013) for DCD. Age at diagnosis of DCD ranged from 4 years 10 months to 8 years 9 months. In Brescia, all the children consecutively referred to the Unit of Child Neurology and Psychiatry of ASST Spedali Civili of Brescia between October 2016 and October 2019 for a suspected diagnosis of DCD were included in the study if they met DSM-5 criteria for a diagnosis of DCD. Age at diagnosis of DCD therefore corresponded to age of enrolling in the Brescia group.

Typically Developing Children (Typically Developing Control Group)

TD children in the Control group were recruited in three United Kingdom schools. Parents and guardians were invited to give consent for their children's participation. Consent was given for 87 children, but two children were excluded due to teacher reports of a diagnosis of ADHD. Following assessment, 16 further children were excluded from the sample for performance below the 15th percentile on the M-ABC (Henderson and Sugden, 1992) or the British Picture Vocabulary Scale (BPVS-II; Dunn et al., 1997). Here we present data from 69 children aged between 5:0 and 12:11 years, to match the age range of the DCD sample. Their

TABLE 1 | Demographic and clinical data of the samples.

Groups	N (M/F)	Age (years) Mean (SD)	Cognitive assessments Standard scores Mean (SD)				Additional diagnoses and clinical data						Movement ABC Scaled scores Mean (SD)			
			BPVS-II	CPM	WPPSI III-WISC IV (Verbal IQ)	WPPSI III-WISC IV (Full IQ)	ADHD	Specific learning disorder	Speech and language disorder	Autism spectrum	Hypermobility	Premature dexterity (<34 Weeks)	Manual dexterity	Aiming and Catching	Balance	Total score
London DCD group	17 (12/5)	9.47 (2.49)	109.4 (17.3)	90.3 (19.7)			1	1	6	1	5	1	4.24 (1.25)	5.35 (3.35)	5.0 (2.69)	3.35 (0.86)
Brescia DCD group	85 (73/12)	8.45 (1.89)			106.8 (15.4)	97.7 (13.7)	2	9	11	0	16	10	5.15 (1.81)	6.09 (2.57)	5.22 (2.26)	4.25 (1.67)
Control group	69 (35/34)	9.12 (1.81)	105.7 (10.71)				0	0	0	0	0	0	≥ 7	≥ 7	≥ 7	≥ 7

Legend: M, male; F, female; sd, standard deviation; BPVS-II, British Picture Vocabulary Scale; CPM, Coloured Progressive Matrices; WPPSI III, Wechsler Preschool and Primary Scale of Intelligence—third version; WISC IV, Wechsler Intelligence scale for children—fourth version; ADHD, Attention Deficit Hyperactivity Disorder.

details are included in **Table 1**. However, it is not meaningful to analyse individual M-ABC scores for this TD group, since the M-ABC is scored such that performance on any item above the 25th centile receives the same score, i.e., data on a TD sample shows a very strong ceiling effect. IQ data (other than receptive vocabulary score) were also not available for the TD group.

Procedures

Assessments of Motor Competency

The M-ABC was used to measure gross and fine motor competency. The London group were tested with the first version of the M-ABC (Henderson and Sugden, 1992) and the Brescia group with the M-ABC-2 (Henderson et al., 2007; Biancotto et al., 2013). Both versions comprise three subsections measuring manual dexterity, aiming and catching, and balance skills. Scores from the three subsections were weighted as specified in the test manual to produce a total impairment score. The M-ABC and M-ABC-2 use different scoring scales, but both are defined in terms of centiles within the population for the age range concerned. For the purpose of this study, both scales were converted into centiles and standard scores to allow analysis of the combined data.

Assessments of Intellectual Ability

The London group of children with DCD were assessed on Raven's Coloured Progressive Matrices (Raven et al., 2008) as a test of fluid IQ, and both children with DCD and TD controls were assessed on the BPVS-II (Dunn et al., 1997). The Brescia DCD group were assessed for both verbal and non-verbal ability using the Wechsler Intelligence scales (WPPSI III = Wechsler Preschool and Primary Scale of Intelligence or WISC IV = Wechsler Intelligence Scale for Children; Wechsler,

2008, 2012). Performance on the WPPSI III or WISC IV and the CPM was used as a measure of full IQ in the present study. Results on these tests of intellectual abilities are included in **Table 1** for information.

Assessment of Global Form and Motion Sensitivity

Versions of the “Ball in the Grass” test developed by Dr. John Wattam-Bell in London (see Braddick et al., 2016) were used in both centres to test children's coherence thresholds for global form (GF) and global motion (GM). Sensitivity to global form and motion was determined by the threshold for detecting global structure as a percentage of coherently organized elements embedded among random noise elements. The test used concentric stimulus displays (Atkinson and Braddick, 2005) which are designed to make the form and motion tasks as comparable as possible, in terms of cognitive demand. Children viewed a laptop computer screen and in each presentation were asked to report whether a circular region—“the ball,” containing concentrically organized short arcs (for GF) or trajectories of moving dots (for GM), was hiding in “the grass” (a background of randomly oriented arcs or randomly directed motion elements) on the left or right of centre (see **Figure 1**). For GM testing, the moving dots had asynchronous limited lifetimes to prevent local tracking and minimise coherent stimulus flicker.

Dimensions of the displays were slightly different for the software versions used in the two studies; details are given in **Table 2**.

Each child in this study completed one run with form and one run with motion. On each trial, the structured target region was presented randomly on the left or right of centre, and

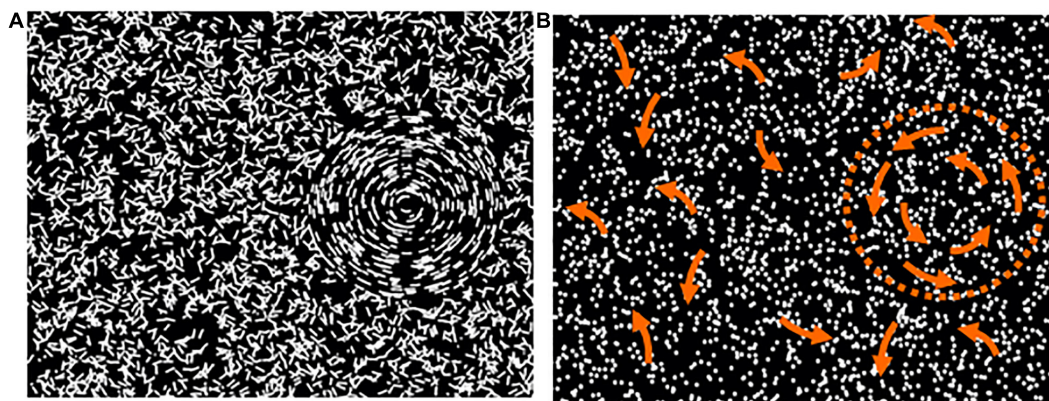


FIGURE 1 | Schematic depiction of examples of the global form (A) and global motion (B) displays. The arrows in B depict the directions of motion inside and outside the marked circular region—neither the arrows nor the dotted circle were visible in the test.

TABLE 2 | Parameters of the global form (GF) and global motion (GM) stimuli used in the Brescia and London studies.

Test	Display size (deg arc) viewed at 50 cm	GM number of dots	GM Dot diameter (min arc)	GM Dot speed (deg/s)	GM Dot lifetime (frames, ms)	GF number of arcs	GF arc size (min)	Target region diameter (deg)	Distance target centre from screen centre (deg)
Brescia	25 × 18	3,000	11	4.1	8, 133	3,000	42 × 8	9.5	6.3
London	32 × 24	3,000	17	4.5	8, 133	2,000	84 × 17	9.5	8.0

the child was asked to point to the side which contained the circular pattern, or for older children to press the corresponding arrow key on the keyboard. Each run began with coherence fixed at 100% with feedback, and these trials were continued until the tester was satisfied that the child understood the task. In the following test phase, the coherence level of the target region was varied without feedback according to the PSI adaptive procedure (Kontsevich and Tyler, 1999) giving an estimate of coherence threshold after the completion of 30 trials. Most children enjoyed the “Ball in the Grass” game and completed testing without difficulty.

Normative Global Form and Global Motion Data

The normative data from earlier samples in London ($N = 184$, Atkinson and Braddick, 2005) and San Diego ($N = 153$, Braddick et al., 2016) were used to derive percentile values and hence scaled scores for GF and GM sensitivity within each 1-year age band in the range of the present samples, with 41–65 children contributing data to each age band. The London sample was tested with one determination of each threshold whereas the San Diego data were based on the mean of two determinations of each threshold for each child. These scaled scores were used in the analyses below of GF and GM sensitivity in relation to M-ABC scores.

The test stimuli used in these normative studies were identical to those used in the Brescia DCD group of the present study. The TD control group was tested in London with the stimulus dimensions as for the London DCD group, i.e., those given in Table 2. Figure 2 plots normative data from the combined London and San Diego samples, and from the present TD control group, showing that the functions relating global form and motion sensitivity to age are aligned for these groups despite the small stimulus differences. A regression analysis (see section

“Statistical Analysis” below) showed no significant effect of group, or interaction of group with age for either GM or GF. It was therefore considered appropriate to use the London TD group as controls for both the London and Brescia DCD groups, and the earlier data set as a basis for the scaled scores.

Statistical Analysis

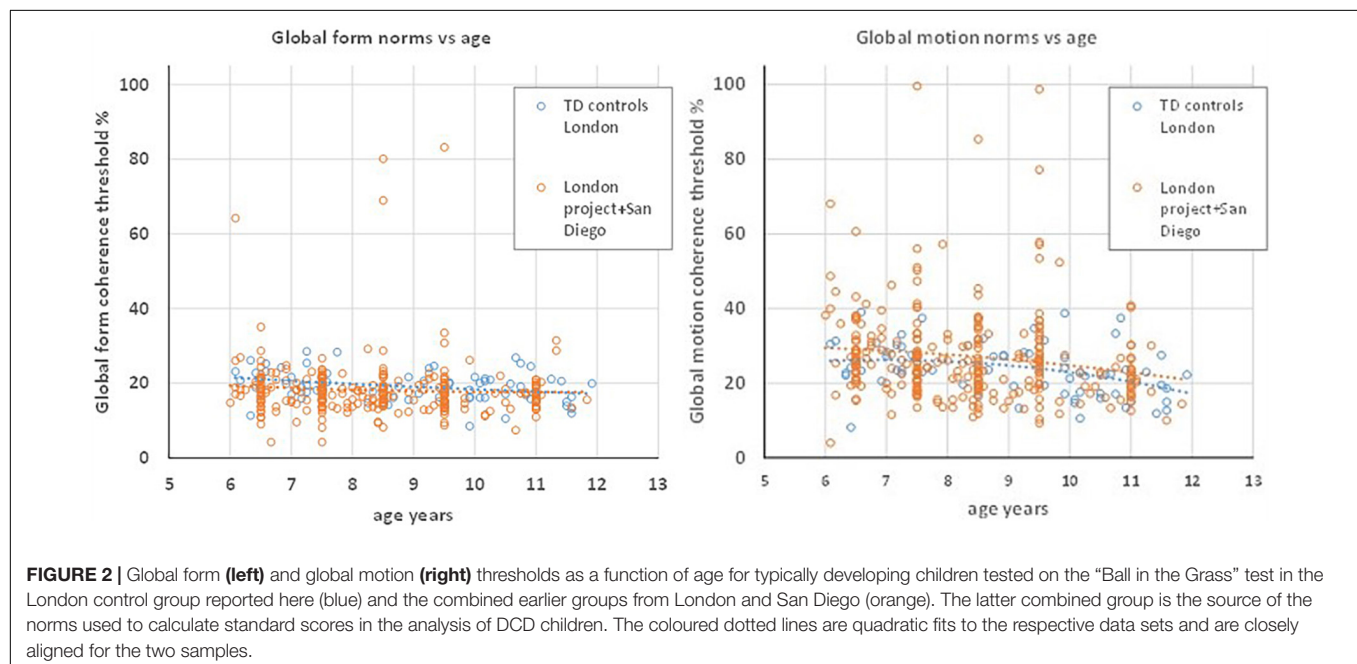
All analyses were performed using R (version 4.0.3; R Core Team, 2016). Continuous data (GM, GF and M-ABC scores) were modelled using ordinary least square regression models. Non-linear trends were modelled using restricted cubic splines with 3 knots. Estimates are reported with associated 95% confidence intervals. All tests were two-sided and assumed a significance level of 5%.

RESULTS

Comparison of Developmental Coordination Disorder and Typically Developing Control Groups

Table 1 shows the descriptive statistics of the DCD group (London Group plus Brescia Group) and the TD control group. We first compared scaled scores for global motion and global form coherence sensitivity between the DCD and TD Control groups in order to examine whether children with DCD showed evidence of dorsal stream vulnerability. Neither age nor gender made a significant contribution to these scores for the two groups together, either on GM (age: $p = 0.441$; gender: $p = 0.805$) or on GF (age: $p = 0.976$; gender: $p = 0.382$).

Compared to the control group, the DCD group showed significantly poorer scaled scores for coherence sensitivity to global motion (“least squares” means adjusted for age and gender



in the model: 8.68 vs. 10.38, delta 1.7, $CI_{95\%}$ 0.64; 2.75, $p = 0.002$), but not to global form (“least squares” means: 8.78 vs. 8.97, delta -0.19 , $CI_{95\%}$ -1.14 ; 0.77, $p = 0.695$). **Figure 3** shows the individual data and linear relationships fitted from the model for each group, and **Figure 4** presents box-and-whisker plots showing the median, quartile and range for each of these data sets.

In the DCD group, full scale IQ (FIQ) was not associated with global motion coherence sensitivity ($p = 0.39$) while better global form coherence sensitivity was associated with a higher FIQ ($p = 0.006$). This pattern was also replicated in the results from the WISC IV (Brescia DCD group only; GM: $r = 0.17$, $p = 0.23$ and GF: $r = 0.28$, $p = 0.02$). Overall, global form and global motion were significantly correlated ($r = 0.29$, $df = 169$, $p < 0.001$), as previously reported by Braddick et al. (2016).

Relationship of Movement-ABC Scores to Global Form and Motion in the Developmental Coordination Disorder Group

In order to examine the relationship of GM and GF sensitivity to the level of motor deficit of individuals within the DCD group, the association of GM and GF scaled scores to the total M-ABC standard scores was analysed, in a regression model including FIQ scores and age. The same analysis was carried out with each of the subsection scores (Manual Dexterity, Aiming and Catching, and Balance). **Figure 5** shows the relationship between total M-ABC scores and GF and GM scaled scores. Total M-ABC standard scores showed a significant linear relationship with global form coherence sensitivity ($p < 0.001$).

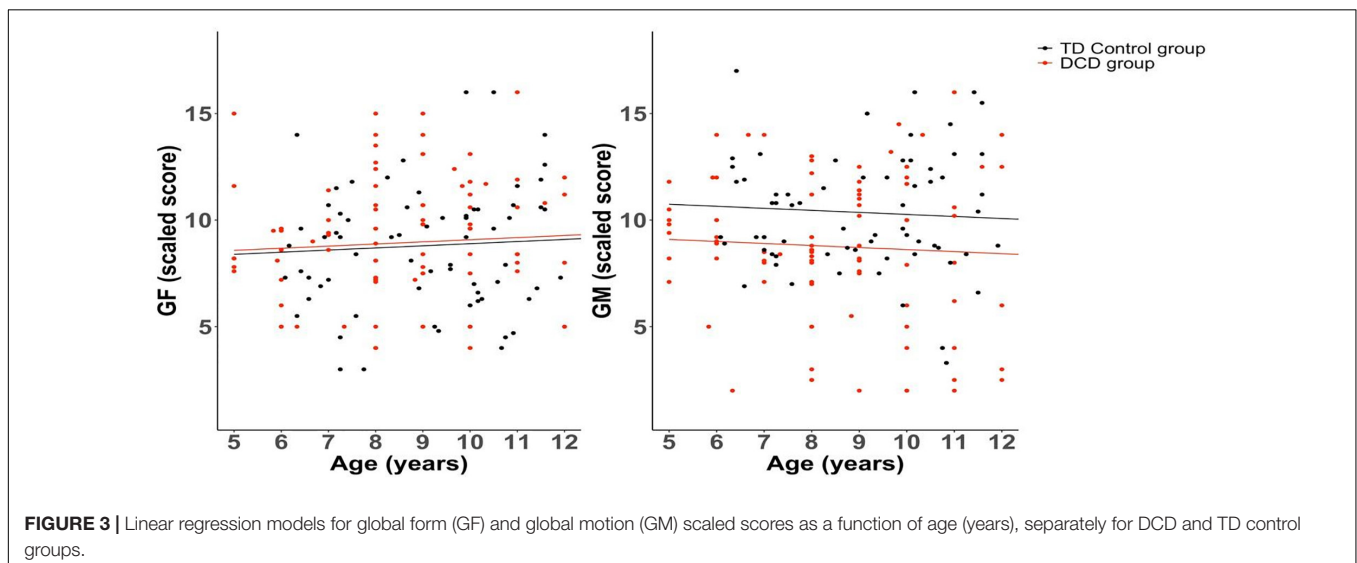
The significant quadratic relationship with global motion coherence sensitivity ($p = 0.046$) was more complex. As shown in **Figure 5B**, low values of GM scaled scores are unrelated to M-ABC, but M-ABC increases over the high range of GM scaled scores. This relationship is not age-dependent ($p = 0.329$) and is not influenced by full IQ test scores ($p = 0.326$).

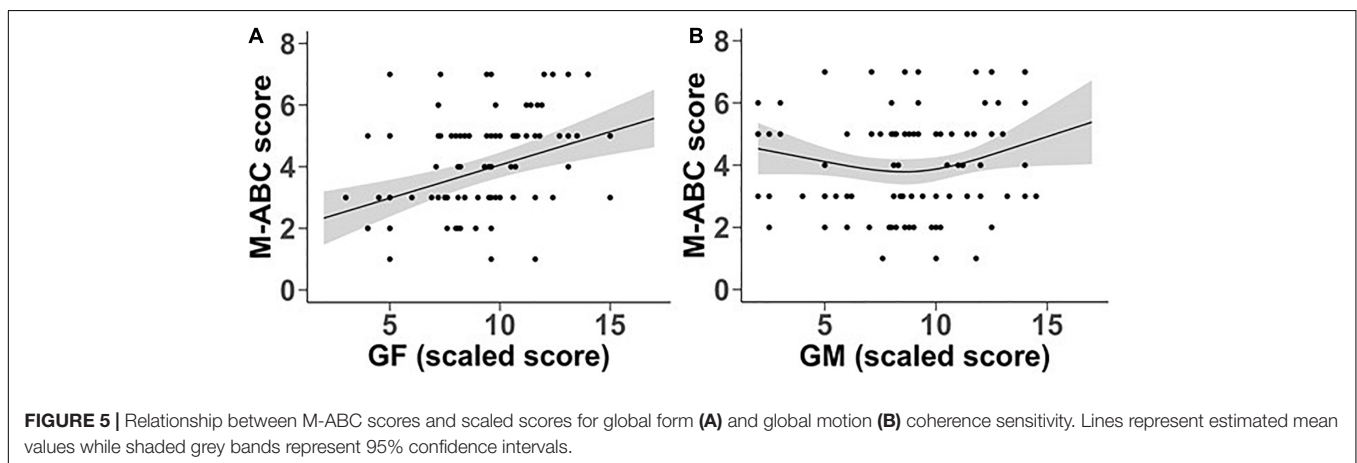
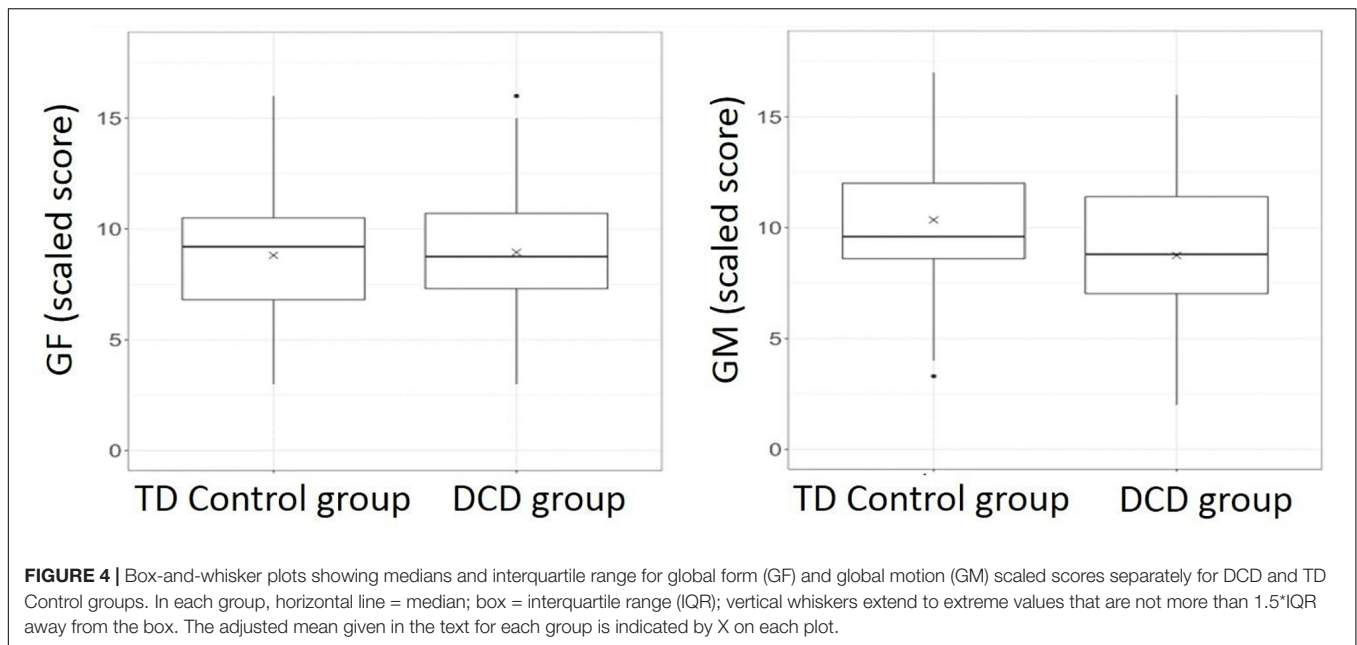
Examination of scores for each M-ABC subsection showed that global form coherence sensitivity was significantly linearly related to Aiming and Catching ($p = 0.003$) and Balance ($p = 0.001$), with the relationship between Manual Dexterity and global form coherence sensitivity only marginally significant ($p = 0.07$). There was no significant linear relationship between scores for each M-ABC subsection and global motion coherence sensitivity (Manual Dexterity $p = 0.370$; Aiming and Catching $p = 0.639$; Balance $p = 0.987$).

DISCUSSION

As discussed in the Introduction, previous studies comparing children with DCD to TD controls yielded conflicting results. The present study used a much larger sample of children, uniformly confirmed as having DCD both by DSM-5 criteria and by results on a standardized battery (M-ABC) and allowed variation of global form and motion sensitivity with age to be taken into account. The contrast between the sensitivity to global form and motion, from two parts of the “Ball in the Grass” test designed to be closely similar in their general cognitive demands, provides some confidence that the difference between children with DCD and TD controls is not a consequence of any general difference between the groups.

However, the results within the DCD group indicate that the relationship between motor skill deficits and global visual perception is more complex than indicated from the comparison with TD controls. In children with DCD, with varying levels of motor skill deficits, motor performance as assessed by the M-ABC showed a significant linear association with global form sensitivity even when age and IQ effects were taken out in the regression model. Global motion sensitivity showed no such linear relationship. However, the significant quadratic relationship seen in **Figure 5** indicates that over the lower range of global motion sensitivity, M-ABC scores showed no systematic relationship with global motion sensitivity, but that at median





levels of global motion sensitivity and above, higher motion sensitivity was associated with higher levels of motor skill.

Thus, in response to the questions posed in the Introduction, we find (a) that motor skill performance is related to both global form and global motion sensitivity, and this relationship is unrelated to either (b) age or (c) IQ measures. It should be noted that sensitivity measures for global motion and global form are significantly correlated with each other. However, the relationships to motor skill illustrated in **Figure 5** come from a regression model in which both form sensitivity and motion sensitivity are entered. Therefore, the distinctive patterns of these relationships to motor skill must reflect the contribution of parts of the variance that are unique to global form and global motion sensitivity, respectively. Neither relationship shows a variation with age, and the results of including IQ in the regression model indicate that the associations do not simply reflect general cognitive ability.

A possible interpretation of these results is that two separate visual components are linked to Developmental Coordination

Disorder. The first of these visual components is associated with global form sensitivity and acts uniformly on the range of motor impairment seen in DCD but has little impact over the higher range of motor performance which differentiates typically developing controls from children diagnosed with DCD. It should be noted that global form sensitivity also showed a linear association with IQ in this population.

A second visual component, associated with global motion sensitivity, only starts to contribute when it reaches a relatively high level in this cohort, where it begins to be associated with higher levels of motor skills. This second component is the dominant factor associated with the much higher levels of motor skill performance which differentiate typically developing controls from children diagnosed with DCD.

Global motion is processed in brain areas within dorsal stream networks such as the intraparietal sulcus (Sunaert et al., 1999; Braddick et al., 2001; Helfrich et al., 2013) and has been taken as a functional signature of the dorsal cortical stream. The dorsal stream is known to be important for sensory-motor

transformations (Buneo and Andersen, 2006; Kravitz et al., 2011). These sensory-motor transformations are essential for the motor skills tested by the M-ABC: spatial vision is required for fine manual control tasks such as bead threading and placing pegs in holes; visual motion processing is key for ball skills (Regan, 1997) and dynamic balance (Sparto et al., 2006). The relationship of these skills to global motion processing is therefore likely to reflect a shared basis in the structure and function in the dorsal cortical stream, and its vulnerability to adverse conditions in development. However, it is only the higher levels of motion sensitivity which reflect this shared basis; when global motion sensitivity is at the lowest levels, other factors appear to dominate in determining the level of motor skill.

A neural interpretation of the linear relationship of global form sensitivity with the lower range of motor abilities within the DCD group is less clear. Low levels of motor skills appear to be related to form sensitivity, but at the levels which differentiate typical development from DCD, form sensitivity shows no association with motor skills. Further research and analysis may yield more insight into what aspects of children's motor coordination are linked to visual dorsal and ventral stream performance, respectively. It should also be noted that dorsal and ventral processing are not independent of each other; the two streams are linked by the vertical occipital fasciculus (Yeatman et al., 2014) and in other connections. The role of these connections in the development of skilled motor behaviour in children is yet to be explored.

Limitations

It was not possible to examine the relationship between motor performance and global form and motion sensitivity in the typically developing control group, since the M-ABC is scored to differentiate between children's motor skills within the low end of the performance range and therefore will not represent variations in motor skills in the typical control group. The presented data are derived from two samples which differed in their recruitment method, which may be responsible for the difference in overall level of impairment apparent in **Table 1**; it is plausible that the London children whose families participated in the Dyspraxia Foundation, and who were overall somewhat older, had more severe impairment than the younger, suspected cases who formed the Italian sample. There were also small differences in stimulus parameters as described above and the version of the M-ABC battery used. However, (a) the stimulus differences do not appear to lead to any systematic differences in performance of typically developing children (**Figure 2**); (b) the differences between M-ABC and M-ABC-2 are primarily in the scoring system, which have been handled here by deriving centiles and hence standard scores from each version of the battery, allowing a unified data presentation. Both samples met the same criteria in terms of their M-ABC performance, and their inclusion in a common analysis adds generality and strength to our conclusions.

CONCLUSION

The present study of a large, well-characterised group of children diagnosed with Developmental Coordination Disorder shows

that the presence and level of their deficit in motor skills have clear associations with global visual coherence sensitivity to both static form and motion. The deficit in global motion sensitivity is shared with a range of other neurodevelopmental disorders and is expected from the known sensory-motor functions of networks in the dorsal cortical stream. However, when a marked deficit of global form processing is present, this dominates the association between visual perceptual and motor skill impairments. This latter relationship will hopefully be pursued further in future research at both neural and functional levels to enable effective interventions.

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 Comitato Etico di Brescia, Brescia, Italy (NP 3513) and UCL Ethics Committee, London, United Kingdom (2807/002). Written informed consent to participate in this study was provided by the participants' legal guardian/next of kin.

AUTHOR CONTRIBUTIONS

SM and FC: study design and conception, data collection, data interpretation, manuscript writing, and critical revision of the article. JA and OB: design and provision of test material, advice on study design and interpretation, part drafting, and critical revision of manuscript. PM and JG: data collection and critical revision of the article. SC: data analysis and interpretation, critical revision of the article. EF: study design and conception, data interpretation, and critical revision of the article. All authors provided final approval of the version to be published.

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