

Effect of comorbid developmental dyslexia on oculomotor behavior in children with developmental coordination disorder: A study with the Developmental Eye Movement test

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- 3 Effect of comorbid Developmental Dyslexia on oculomotor behavior in children with
- 4 Developmental Coordination Disorder: A study with the Developmental Eye Movement test

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Abstract

Studies have suggested a dysfunction in oculomotor skills in children with developmental coordination disorder (DCD). It has been proposed that the Developmental Eye Movement (DEM) test is useful in testing the dyslexics' (DD) oculomotor behavior during reading, in a simple and indirect manner. The present study aimed at exploring the oculomotor behavior in children with DCD as assessed with the DEM test. Furthermore, we compared children with DCD to children with DD and to children with both DCD and DD in order to investigate the specificity of the oculomotor difficulties, as measured by the DEM test. Results showed that 1) children with DCD presented mild atypical performances at the DEM test (error z-score only), 2) children with DD presented particularly poor performance on the DEM test, and 3) the co-morbid condition (DCD+DD) did not add to the severity of atypical performance obtained on the DEM test. In sum, children with DCD were the less affected according to the DEM test, and children with DD (isolated or comorbid) presented the most atypical performance. Results at the DEM test did not allow making emerge clear oculomotor atypicalities in DCD. We thus conclude that more research using eye-tracking techniques is needed to explore the nature of oculomotor atypicalities in DCD children, to distinguish DD and DCD oculomotor behavior, and to understand the profile of children with dual diagnosis.

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Keywords: DCD; Developmental Dyslexia; DEM test; reading; comorbidity; oculomotor behavior

Introduction

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Developmental Coordination Disorder (DCD) is a neurodevelopmental disorder characterized by significant difficulties with the acquisition and execution of motor skill [DSM-5, American Psychiatric Association (APA), 2013]. Individuals with DCD demonstrate a level of motor skill out of keeping with their age and intellectual ability, in the absence of other medical condition (Cerebral Palsy, sensory deficit, etc.). A meta-analysis showed a pattern of deficits which are characteristic of DCD and involving different areas such as internal (forward) modelling, rhythmic coordination, executive functions, gait and postural control, catching and interceptive action, and aspects of sensori-perceptual processing (Wilson, Ruddock, Smits-Engelsman, Polatajko, & Blank, 2013). Information processing has been shown to be altered in this developmental disorder (Wilson and McKenzie, 1998). In particular, the authors identified increased deficits with visuo-spatial processing tasks in the DCD population, regardless of whether or not the tasks involved a motor component. Later, this last result has been confirmed by several studies (Bellocchi, Muneaux, Huau, Lévêque, Jover, & Ducrot, 2017; Beery & Beery, 2004; Biotteau, Albaret, Lelong, & Chaix, 2017; Gardner, 1982; Martin, 2006; Prunty, Barnett, Wilmut, & Plumb, 2016). Additionally, there is an increasing interest in literature over the last decades for eyemovement behavior in DCD while executing various tasks (Gaymard et al., 2017). Some studies showed difficulties in pursuit tracking tasks in individuals with DCD, but the analysis of the results also demonstrated preserved aspects (Langaas, Mon-Williams, Wann, Pascal, & Thompson, 1998; Lord & Hulme, 1988; Robert, Ingster-Moati, Albuisson, Cabrol, Golse, & Vaivre-Douret, 2014; Sumner, Hutton, Kuhn, & Hill, 2016). In particular, Langaas et al. (1998) reported a reduced gain in pursuit eye movements. More recently, Licari et al., (2018) found that DCD individuals took longer to initiate smooth pursuit but once initiated, these individuals were

effectively able to maintain it. Furthermore, Robert et al. (2014) suggested a delay in the maturation of the ocular pursuit system in children with DCD. They compared horizontal and vertical pursuit eye movements of children with DCD with those of TD children and showed that only vertical pursuit gain was significantly lower in children with DCD. In addition to horizontal smooth pursuit, Sumner et al. (2016) examined fixation stability and performance on pro- and anti-saccade tasks in children with DCD. In line with Robert et al. (2014), they showed that horizontal pursuit gains were comparable in DCD and TD children. Moreover, response preparation in the pro- and anti-saccade tasks were not impaired in children with DCD. The authors concluded to intact fundamental neural mechanisms underlying pursuit and saccades in DCD. However, children with DCD had deficits in maintaining engagement in the fixation and pursuit tasks, and have problems with saccadic inhibition compared to TD controls of the same age. Therefore, children with DCD would show oculomotor 'atypicalities' (Sumner et al., 2016)¹.

The eye-tracking technique has also been used to highlight difficulties in eye-hand coordination in DCD. Langaas et al. (1998) showed that DCD children had difficulties in smoothly synchronizing their eye movements with a moving object. Accordingly, Wilmut, Wann and Brown (2006) observed that children with DCD have difficulties in concatenating the sequential shifts of gaze and hand in order to complete everyday tasks or assessment items (i.e., double-step pointing paradigm; see also Katschmarsky, Cairney, Maruff, Wilson, & Currie, 2000, for similar results). Similarly, DCD children were found to have slower strategy of fixating the target prior to initiating a hand movement (Wilmut & Wann, 2008). In the same vein, a

¹ Since a huge variety of eye trackers exists, the differences pointed out by the studies could be due to the different characteristics of the eye-tracking system employed.

group-based gaze training intervention has shown to enhance DCD children's general coordination and perception of their catching ability (Wood et al., 2017). Finally, Warlop and colleagues (2020) recently found differences in gaze behavior between young adults with DCD and typically developing individuals in an everyday life task, i.e. walking.

As a whole, despite some preserved aspects, a large number of studies concluded on the presence of oculomotor 'atypicalities' in children with DCD.

Even though there is a non-negligible amount of studies investigating DCD individuals' eye-movements, the exploration of oculomotor control during reading in that specific population is quite scarce in the literature. Nonetheless, reading difficulties have been observed in 29% to 70% of children diagnosed with DCD (O'Hare & Khalid, 2002). In this context, Bellocchi and colleagues (2015) investigated the landing position pattern in oculomotor control that take place in visual word perception in children with DCD. The authors asked children to perform an oculomotor lateralized bisection task on words, strings of hashes and lines. More precisely, the children were asked to move their eyes as quickly as possible to a position they thought to be the middle of the stimulus and to validate this position by pressing a button. Even if the oculomotor bisection task is not directly a reading task, it elicits saccade targeting which has been proved to be an integral part of the reading process (Ducrot & Pynte, 2002; see Bellocchi, Massendari, Grainger, & Ducrot, 2019 for data on Developmental Dyslexia -DD²). Results showed that

² Developmental dyslexia (DD) is a neurodevelopmental disorder that is diagnosed when no sensory and intellectual deficits can explain reading and/or writing disorders and when adequate instruction and socio-cultural opportunities are available but fail to result in an adequate level of performance [DSM-5, American Psychiatric Association (APA), 2013; W.H.O., 1992].

children with DCD had slower saccade latency compared to TD children in programming their first saccade and that they were less accurate on saccade targeting. These difficulties particularly emerged while they had to move their eyes on stimuli presented in the left-visual field (LVF) which requires programming a right-to-left saccade. These results suggested less automatized procedures for saccade targeting in children with DCD.

To our knowledge, it is unknown whether the oculomotor control atypicalities found in DCD may be explained or influenced by the presence of a co-occurring neurodevelopmental disorder (see Smits-Engelsman, Jover, Green, Ferguson, & Wilson, 2017). Indeed, co-occurrence between neurodevelopmental disorders is a very common condition. Because DCD often co-occurs with DD, eye movement disorders in this population could rely on reading subclinical deficits. Indeed, epidemiological studies demonstrate a rate of comorbid diagnosis of DCD and DD in 16% (Kaplan, Dewey, Crawford, & Wilson, 2001) to 70% of DCD children (Iversen, Berg, Ellertsen, & Tønnessen, 2005). Similarly, Chaix and colleagues (2007) found an high percentage of DCD diagnosis in a group of 58 dyslexics, i.e. 40% scored below –2 standard deviations (SDs) on the Lincoln-Oseretsky Motor Development Scale (Rogé, 1984), and 17.2% scored between –1 and –2 SDs.

Unfortunately, despite the evidence of a frequent association between DCD and DD, this co-occurrence has not been taken into consideration in the research exploring oculomotor control in these two clinical populations (i.e., DCD and DD). Bellocchi and colleagues (2015) did not observe any difference between "DCD+DD" and "isolated DCD" children in the oculomotor bisection task. However, DCD+DD children differed from DD children, suggesting that the presence of a motor disorder might induce a dysfunction in saccade programming. Beyond the question of oculomotor control, recent studies exploring the impact of the co-occurrence between DCD and DD on visuo-spatial abilities, have pointed out the absence of significant differences

Perception and Visual-Motor Integration quotient components of the DTVP-2 (Bellocchi et al., 2017), but also in visuo-attentional processes involved in word recognition (i.e., *Optimal Viewing Position*³ - *OVP*- Bellocchi & Ducrot, submitted). As a whole, these data suggest that the comorbid condition does not sharpen difficulties in eye movements or in visuo-spatial processing in DCD children. Previous studies have reached the same conclusion, with no cumulative impact on cognitive abilities in the case of a dual diagnosis (see Biotteau et al., 2017, for a detailed review; Kaplan, Crawford, Cantell, Kooistra, & Dewey, 2006). However, recent findings depict a more complex framework than what it looks like. Indeed, Bellocchi and Ducrot (under review) suggested that children with dual diagnosis show an intermediate profile between children with isolated disorders [see also Maziero, Tallet, Bellocchi, Jover, Chaix & Jucla (2020), for similar results on working memory capacity]. It is clear that this point deserves a particular attention in scientific studies. We will thus try to address that question here.

The present study

Despite the evidence of 1) the importance of the oculomotor control on different cognitive tasks, and 2) data showing an oculomotor dysfunction in DCD, the assessment of eye movements is not systematically carried out and widespread in the clinical practice and the consideration of the comorbidity with dyslexia is neglected (see Bellocchi, Muneaux, Bastien-Toniazzo, & Ducrot, 2013 for a review on DD). One of the reasons resides on the complexity of the technical

³ The *Optimal Viewing Position (OVP)* effect indicates that words are identified most quickly when the eyes fixate near the word centre in alphabetic languages (e.g., Farid & Grainger, 1996; Nazir, O'Regan, & Jacobs, 1991; O'Regan & Jacobs, 1992; Vitu, O'Regan, & Mittau, 1990).

apparatus needed to record on-line oculomotor control during the execution of a cognitive task. A response to this limit has been represented by the Developmental Eye Movement (DEM) test (Garzia, Richman, Nicholson, & Gaines, 1990) that provides an indirect measure of oculomotor abilities involved in a rapid automatized naming (RAN) task. It comprises horizontal and vertical digit naming tasks and provides reading time and errors norms. Thus, it is a simple and easy visual-verbal paper-based test used as an oculomotor assessment tool in optometric clinical practice guideline (e.g., Facchin, Maffioletti, & Carnevali, 2011; Facchin, Ruffino, Facoetti, & Maffioletti, 2014; Medland, Walter, & Woodhouse, 2010; Tassinari & DeLand, 2005). Another advantage is that the DEM test provides four behavior profiles, which can disentangle RAN difficulties from oculomotor dysfunction: 1) Type 1, which represents normal automaticity of digit naming and oculomotor skills. 2) Type 2, which represents oculomotor dysfunction. 3) Type 3, which represents difficulties in automaticity of digit naming. 4) Type 4, which is a combination of Type 2 and 3 and represents difficulties in digit naming and oculomotor skills. In order to validate the structure of the DEM test, Facchin et al. (2011) revealed that the results of a factorial analysis showed saturation to three main factors. The first factor was linked to the common naming component between vertical and adjusted horizontal time. This factor has been thus referred to automatized naming skills. The second factor was related to adjusted horizontal time and Ratio (i.e. the quotient between horizontal time and vertical time), and it has been referred to oculomotor skills. They finally found a third factor constituted by Errors. These three factors account for 99.8% of the total variance of the DEM test data. The authors concluded that the results of the factorial analysis clearly differentiated naming skills from oculomotor skills and errors. Accordingly, atypical scores obtained in the first two components could reveal difficulties in naming and oculomotor skills, respectively.

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Despite being criticized (e.g., Ayton, Abel, Fricke, & McBrien, 2009; Webber, Wood, Gole, & Brown, 2011), the DEM test has recently been suggested to be useful in exploring the oculomotor behavior of children with DD during reading. Moiroud, Gerard, Peyre, and Bucci (2018) sought to associate the DEM test outcomes with measures of gaze behavior and showed that children with DD and normal readers of equivalent reading level have significant longer fixation times and take longer to read the horizontal reading task than normal readers of similar chronological age. In addition, they found a correlation between the fixation time and the number of words read in one minute with the total time of the horizontal reading task.

As a first goal, our work aimed at exploring oculomotor behavior in children with DCD as assessed with the DEM test. Particularly, according to the interpretation of the results provided by the DEM test, we predicted that if children with isolated DCD showed oculomotor difficulties, they should mainly exhibit Type 2 profile that represents oculomotor dysfunction. In addition, according to the so-called *phonological hypothesis*⁴ (e.g., Snowling, 2000; 2006; Vellutino & Fletcher, 2005; Vellutino, Fletcher, Snowling, & Scanlon, 2004), children with isolated DD should be mostly linked to Type 3 profile which represents specific difficulties in RAN. Subsequently, children with comorbid disorders (DCD+DD) should be mainly characterized by Type 4 profile, which represents a combination of RAN and oculomotor skills difficulties.

⁴ One of the most widely accepted explanations of DD posits a core deficit at the phonological level of processing. In particular, the so-called *phonological theory* asserts that dyslexics have a specific impairment in the representation, storage, and/or retrieval of speech sounds that prevents the proper acquisition of the grapheme-phoneme correspondence necessary for learning to read in an alphabetic system.

Secondly, we wanted to explore the impact of comorbidity on DCD's oculomotor behavior. To this aim, we compared children with DD and DCD (DD+DCD) to children with isolated disorders (DD or DCD) in the DEM task. According to a cumulative hypothesis (e.g. Pitcher, Piek, & Barrett, 2002), if co-morbid condition adds to the severity of the cognitive deficit, children with DD and DCD should display more marked oculomotor atypicalities than children with isolated disorders. On the contrary, if co-morbid condition does not add to the severity of the cognitive deficit, we should find that no difference between children with isolated disorders and those with a dual diagnosis (e.g., Bellocchi et al., 2015; Bellocchi et al., 2017; Biotteau et al., 2017). Moreover, the comparison between co-morbid children (DD+DCD) and children with isolated disorders (DD or DCD) can provide evidences about the specificity of the oculomotor difficulties, as measured by the DEM test, to reading or motor impairments. In other words, if atypicalities at the DEM test are associated with reading deficit, we should find them more specifically in children with reading impairments (isolated DD and DD+DCD). However, if atypicalities at the DEM test are associated with motor disorder, we should find them more specifically in DCD children (isolated DCD and DD+DCD).

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Method

Participants. In total, 138 children (57 girls, 81 boys) participated in the study. Four groups were constituted (see Table 1), one composed of 42 typically developing children (TD), the other composed of 22 children with developmental coordination disorder (DCD), the third composed of 47 children with developmental dyslexia (DD), and the last one composed of 27 children with developmental dyslexia and developmental coordination disorder (DD+DCD). The patients studied were enrolled in the DYSTAC MAP cohort (ANR-13-APPR-0010).

All children were aged between 7 years 8 months to 12 years 6 months, right-handed, and French native speakers. All parents reported a normal hearing, and normal or corrected-to-normal vision of their child. All underwent a full medical and neuropsychological assessment conducted by skilled practitioners. They all had normal intellectual functioning level (standard score at least above 7 at the Similarities and Pictures Concepts subtest, French-language version of the WISC-IV, Wechsler, 2005). Furthermore, children whose oral language skills were in the pathological range (scores below -2 SD on the « EVAC » -Flessas & Lussier, 2003- and « Ecosse » -Lecocq, 1996- tests), or who were diagnosed with ADHD (six or more symptoms of the « Hyperactivity/impulsiveness » as « Inattention » checklist of the DSM-5) were excluded from the study.

[Insert Table 1 about here]

Group	TD	DCD	DD	DCD+DD	Group diff	Post hoc comparison
N (females)	42 (20)	22 (7)	47 (20)	27 (10)	ns	
Age (M ±SD)	$10,0 \pm 1,1$	$9,8 \pm 1,3$	$10,1 \pm 1,1$	$10,1 \pm 1,3$	ns	
DSM-5 ADHD questionnaire total score (M ±SD)	$0.86 \pm 1,2$	5,41 ±2,6	4,36 ±2,4	5,36 ±2,7	<.001	TD>DD;DCD;DCD+DD
MABC total score (percentiles) (M ±SD)	$4,06 \pm 3,1$	20,84 ±4,8	5,45 ±2,7	$20,33 \pm 5,3$	<.001	TD;DD>DCD;DCD+DD
Alouette Accuracy Z-score (M ±SD)	0,57 ±0,3	$0,32 \pm 0,4$	-2,19 ±1,2	-2,24 ±1,9	<.001	TD>DCD>DD;DCD+DD

Alouette Speed Z- score (M ±SD)	$0,95 \pm 0,8$	$0,31 \pm 0,8$	-1,43 ±0,6	-1,35 ±0,5	<.001	TD>DCD>DD;DCD+DD
Odedys Irregular word Accuracy (M ±SD)	$0,84 \pm 0,6$	$0,63 \pm 0,7$	-1,63 ±1	-1,93 ±1,1	<.001	TD;DCD>DD;DCD+DD
Odedys Irregular word Speed (M ±SD)	$0,80 \pm 0,4$	0,54 ±0,5	-2,92 ±3,5	-1,94 ±2,1	<.001	TD;DCD>DD;DCD+DD
Odedys Pseudo word Accuracy (M ±SD)	0,51 ±0,6	0,03 ±0,5	-1,8 ±1,3	-1,63 ±1,1	<.001	TD>DCD>DD;DCD+DD
Odedys Pseudo word Speed (M ±SD)	0,54 ±0,6	0,25 ±0,6	-2,17 ±3,3	-1,11 ±1,2	<.001	TD;DCD>DD;DCD+DD

Table 1: Demographic characteristics and inclusion criteria of each group of children. Sex ratio, age, DSM-5 ADHD questionnaire total score, MABC total score (percentiles), Alouette Accuracy and Speed z scores and Odedys Accuracy and Speed z scores for Irregular and Pseudo word reading. Note. TD = typically developing children; DCD= children with isolated DCD; DCD+DD= children with both DCD and DD; Group diff= Group difference. Group differences were tested with a Kruskall Wallis test and χ^2 (sex ratio), and post-hoc test with Dwass-Steel-Critchlow-Fligner test.

Participants with diagnosed neurodevelopmental disorders were recruited at San Salvator University Hospital [Center for the diagnosis of learning disabilities] in Marseille, at the Hospital of Aix-en-Provence, and at the Purpan University Hospital in Toulouse, France. Children with DCD were included in the study if they had a score below the 5th percentile on the French version of the M-ABC (Soppelsa & Albaret, 2004). Children with DD were included in the study if they showed manifested reading deficits in two different tests: at least 1 SDs below the normal level on the "Alouette Test-R" (Lefavrais, 2005), a test accounting for speed and accuracy of text reading, and at least 1.5 SDs below the normal level on the ODEDYS-2 test- Outil de dépistage

des dyslexies, Second Edition (Jacquier-Roux, Valdois, & Zorman, 2002), a test assessing speed and accuracy of reading isolated irregular words and pseudowords.

Children were included in the group of children with isolated DCD only if they had no reading difficulties [at least -0.5 SDs above the normal level on the "Alouette Test-R" (Lefavrais, 2005) and on the ODEDYS-2 test- Outil de dépistage des dyslexies, Second Edition (Jacquier-Roux et al., 2002)]. Children were in the group of children with isolated DD only if they had no motor difficulties [scores above the 15th percentile on the M-ABC test]. Children of the group DCD+DD had a score below the 5th percentile on the M-ABC and reading impairments [i.e., reading performance respecting the two previously presented criteria - at least 1 SDs below the normal level on the "Alouette Test-R" and at least 1.5 SDs below the normal level on the ODEDYS-2 test].

Finally, children in the TD group were recruited through announcement in schools and in the laboratories. They had normal reading abilities [at least -0.5 SDs above the normal level on the "Alouette Test-R" and on the ODEDYS-2 test] and normal motor abilities [scores above the 15th percentile on the M-ABC test]. None of them suffered from any neurological, psychiatric, or emotional disorders or were educationally disadvantaged. We did not include children who were considered by parents or by the practitioners' assessment as having either a specific learning deficit or cognitive and behavioral problems.

This work was conducted in accordance with the Declaration of Helsinki (WHO, 2008), approved by the French Ethics Committee Review Board (2014-A01239-38 and 2014-A01960-47). The children and their parents gave their written consent for participation.

Materials. Each participant was given the DEM test (Garzia, et al., 1990) which is composed by horizontal and vertical digit reading tasks printed on four different sheets of paper:

the pre-test (a horizontal line of ten 0,5 cm high digits), two vertical tests (Test A and B; each composed of two vertical lines of twenty 0,5 cm high digits separated by a 10.5 cm horizontal margin and with a 0,5 cm vertical distance between letters) and one horizontal test (Test C; sixteen lines of five irregularly separated 0,5 cm high digits). Children were asked to read aloud the digits as fast and as accurately as possible. The task was timed, and errors or omissions were recorded. The DEM test provides several measures: 1) the vertical reading time (seconds) (VT) which represents the sum of the time spent on naming the eighty vertically organized digits of Test A and B (in accordance with the test manual, errors were not used for scoring purpose); 2) the adjusted horizontal time (second) (HT) which represents the time required for reading the eighty horizontally organized digits presented in the Test C; this score is corrected for omission or addition errors⁵; 3) the total number of errors which gives the accuracy on the execution of Test C (errors recorded: omission, addition, substitution, transposition); 4) the Ratio score which is calculated dividing the HT by the VT.

Furthermore, for clinical purposes, the DEM test provides four behavior profiles: 1) Type 1, which represent normal automaticity of digit naming and oculomotor skills. It comprises essentially normal performance in HT, VT and Ratio. The children were included in this group if all the scores were > -1.5 SD. 2) Type 2, which represents oculomotor dysfunction. It is characterized by abnormally increased scores in HT and Ratio but normal scores for VT. The children were included in this group if HT and Ratio scores were <= -1.5 SD and VT scores > -1.5 SD. 3) Type 3, which represents difficulties in automaticity of digit naming. It comprises abnormally increased scores in VT and HT, but normal scores in Ratio. The children were included in this group if VT and HT scores were <= -1.5 SD and Ratio scores > -1.5 SD. 4) Type

⁵ Adjusted Horizontal Time = Test C time x [80/(80-number of omission + number of addition)]

4, is a combination of Type 2 and 3 and represents difficulties in digit naming and oculomotor skills. It is characterized by abnormally increased scores in VT, HT and Ratio. The children were included in this group if all the scores were <= -1.5 SD.

Procedure. All children were tested individually at the hospital and at the university laboratories by a neuropsychologist and a psychomotor therapist enrolled in the research project for their expertise in neurodevelopmental disorders. All the children received a comprehensive cognitive assessment to evaluate criteria for inclusion in each group. This session lasted about 1 hour and a half. However, the administration of the DEM test lasted about 10 minutes.

Data Analysis. Data were analysed using the Jamovi statistical computer software [The jamovi project (2019). jamovi (Version 0.9). Retrieved from https://www.jamovi.org]. As Shapiro-Wilk test demonstrated that the data were not normally distributed, and the group size were small, all statistics were non-parametric. Group comparisons for the children's test scores were performed using Kruskal Wallis test a non-parametric version of one-way ANOVA on ranks which is usually used for comparing more than 2 independent samples. Post-hoc comparisons were done with Dwass-Steel-Critchlow-Fligner test which compares all pairs of groups using a pairwise ranking nonparametric method and controls the error rate. The distribution of the sex ratio and of the DEM types between the groups were tested using χ_2 tests and pairwise comparisons were performed with Bonferroni correction (p = .008). The significance threshold was set at p = .05.

Results

Kruskall-Wallis test showed a significant effect of group for all z-scores (VT, H(3) = 28.38; p < .001; HT, H(3) = 38.85; p < .001; Ratio, H(3) = 9.19; p < .05; Errors , H(3) = 20.84; p < .001; see Figure 1). Dwass-Steel-Critchlow-Fligner pairwise comparisons demonstrated that children with isolated DCD differed from TD children only for the errors' z-score (p < .039). In contrast, children with DD had lower z-scores than TD children for the horizontal time, vertical time and in the errors. Finally, children with DCD+DD had lower z-scores than TD children for the horizontal time, vertical time, errors and the ratio.

[Insert Figure 1 about here]

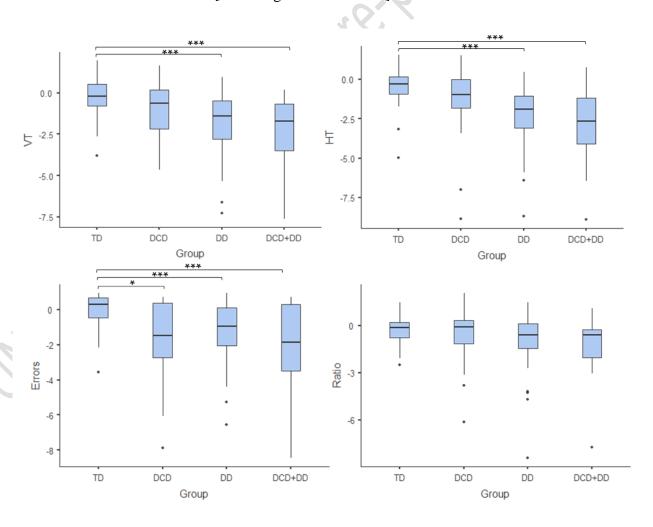


Figure 1: Box plot representation of the DEM variables expressed in z scores. VT: vertical time, HT: horizontal time, Ratio: ratio HT/VT. Note: *p < .05, **p < .01, *** p < .001. Note. TD = typically developing children; DCD= children with isolated DCD; DD= children with isolated DD; DCD+DD= children with both DCD and DD.

The distribution of the behavior types differed significantly between the groups of children (χ_2 = 37.5, p <.001). Pairwise comparisons showed differences between TD and other groups (p <.008) but not between DD, DCD and DCD+DD groups. Children with DCD, as did DD and DCD+DD children, were more often presenting abnormal behavior response type than TD children, but the differences between children with DCD, children with DD and children with DCD+DD were never significant.

[Insert Figure 2 about here]

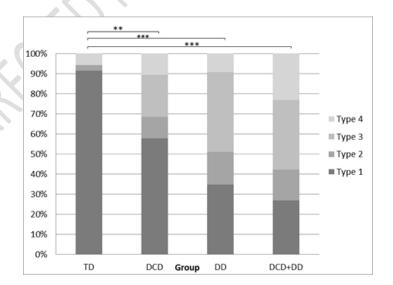


Figure 2: Distribution of the four behavior types in each group. Note: 1) Type 1: normal automaticity and oculomotor skills. 2) Type 2: oculomotor dysfunction. 3) Type 3: difficulties in automaticity of digit naming. 4) Type 4: difficulties in digit naming and oculomotor skills (a combination of Type 2 and 3). TD = typically developing children; DCD= children with isolated DCD; DD= children with isolated DD; DCD+DD= children with both DCD and DD.

Discussion

The first aim of the present study was to explore oculomotor behavior in children with DCD as assessed with the DEM test (Garzia et al., 1990). As a whole, results showed that children with DCD presented slight atypical scores at the DEM test, as attested by the fact that only one parameter was significantly affected. Particularly, children with isolated DCD made more errors if compared to TD children. Children with DCD+DD clearly showed a wider range of difficulties when compared to children with DCD only, and, of course, to TD children: they had lower z-scores than TD children did for the horizontal time, vertical time, errors and the ratio. Additionally, the data revealed that DCD and DCD+DD showed less Type 1, more Type 2, and 4 compared to TD children, thus suggesting atypical profile of oculomotor responses in DCD children (isolated or comorbid)⁶. Contrary to our hypothesis, this result was also observed in children with isolated DD who showed a pattern of deficit similar to children with DCD and DCD+DD. Moreover, no significant difference emerged on the percentage of Type 2 profile

⁶ As a reminder, according to the interpretation provided by the manual of the DEM test, Type 2 and 4 correspond to a wide range of difficulties, going from a specific oculomotor dysfunction (Type 2) to a combination of difficulties in digit naming and oculomotor skills (Type 4).

between the isolated DCD group and the two other groups of neurodevelopmental disorders (DD and DCD+DD), suggesting that isolated DCD group could not be specifically characterized by oculomotor dysfunction as assessed by DEM test.

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Contrary to our expectations, the results of the present study showed that atypical scores at the DEM test were more frequently associated to a diagnosis of DD, both in digit naming and oculomotor skills. As previously described, children with DCD and DD diagnosis, as well as children with isolated DD, showed a wider range of difficulties if compared to TD children. Indeed, they not only made more errors than the DCD only group, but also showed lower scores for the horizontal time, vertical time, ratio and atypical oculomotor responses if compared to TD children (i.e., higher presence of Type 2 and 4). In other words, the results at the DEM test were very sensitive to children's reading abilities or, more specifically, to the presence of a reading disorder. This is what was found by Moiroud and collaborators (2018) who showed that dyslexic children and the reading age-matched children took longer time to read Text C of the DEM test if compared to the chronological age-matched children group. Consistently, in their study, dyslexics and reading age-matched children had longer fixation time compared to chronological agematched children while reading this text. Accordingly, previous important findings showed that compared to age-matched control readers, dyslexics' eye movements in word, pseudoword, or sentence reading are characterized by more and longer fixations. Additionally, dyslexics usually show shorter saccades and more regressions (Bellocchi et al., 2019; Biscaldi, Gezeck, & Stuhr, 1998; Bucci, Nassib, Gerard, Bui-Quoc, & Seassau, 2012; Hawelka, Gagl, & Wimmer, 2010; Hutzler & Wimmer, 2004; McConkie, Zola, Grimes, Kerr, Bryant & Wolff, 1991; Rayner, 1986; Seassau, Gerard, Bui-Quoc, & Bucci, 2014).

The Type 3 profile of the DEM test is supposed to refer to specific difficulties in RAN. In the present study, children with DCD +DD and children with isolated DD tended to be more

linked to this profile, if compared to others profiles and to DCD group. This tendency could be in accordance with the so-called *phonological hypothesis* (e.g., Snowling, 2000; 2006; Vellutino & Fletcher, 2005; Vellutino et al., 2004). Although non-significant, these differences are interesting anyhow because they show a specific pattern of profile regarding children with a DD, associated or not with a DCD. In other words, the DEM test seemed to be sensitive in highlighting RAN deficits in children with reading disorders. These descriptive results deserve to be replicated in future studies.

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Until now very few studies have investigated whether the oculomotor control atypicalities found in DCD may be, or not, influenced by the presence of a co-occurring DD. Considering the evidence of a frequent association between these two neurodevelopmental disorders (Iversen et al., 2005; Kaplan et al., 2001), our second aim here was to explore the impact of comorbidity of DD on DCD's oculomotor skills, particularly the oculomotor skills measured by the DEM test. As a whole, results showed no significant differences between comorbid and isolated groups at the DEM test. This means that no additional impact on DEM performance is associated with a dual diagnosis. Therefore, these data do not support the cumulative hypothesis according to which if co-morbid condition add to the severity of the cognitive deficit, children with DCD and DD should revealed more marked atypical DEM test results than children with isolated disorders (e.g., Pitcher et al., 2002). These results are in line with other recent studies suggesting that the comorbid condition does not systematically add to the severity of associated cognitive disorders (e.g., Bellocchi & Ducrot, submitted; Bellocchi et al., 2017; Biotteau et al., 2017; Kaplan et al., 2006; Maziero et al., 2020). Note that, as previously mentioned, children with DCD and DD diagnosis, showed atypicalities on a wider range of parameters when compared to isolated disorders. Accordingly to what it has been stated above, this result can be mainly explained by the presence of a co-occurred DD.

Even if the differences between the three groups of children with neurodevelopmental disorders (DCD, DCD+DD, and DD) were not statistically significant, it does not mean that these groups had comparable performance on the DEM test. Indeed, if we look at the distribution of the behavior types (Figure 2), it appears that the profiles that emerged are quite heterogeneous.

Particularly, we can observe that DCD+DD group showed higher percentage of Type 4 profile compared to the other two profiles (Type 2 and 3) and compared to isolated DD and DCD. Again, it is important to notice that these differences were not statistically significant but they highlight a variability that researchers need to explore more deeply as it could depict a more complex framework of the DCD+DD comorbid profile.

Finally, results at the DEM test did not reveal clear oculomotor atypicalities in DCD. For instance, the percentage of Type 2 profile, indicating an oculomotor dysfunction, wasn't higher in children presenting a DCD isolated or comorbid, compared to the other neurodevelopmental groups. Rather than concluding that DCD children do not encounter eye-movements difficulties, we assume that the DEM test is not as sensitive to oculomotor than to RAN difficulties. Indeed, first of all, the vast majority of researches observed oculomotor difficulties in DCD (e.g., Bellocchi et al., 2015; Gaymard et al., 2017; Gonzalez et al., 2016; Langaas et al., 1998; Katschmarsky et al., 2000; Licari et al., 2018; Lord & Hulme, 1988; Robert et al., 2014; Sumner, et al. 2016; Warlop et al., 2020; Wilmut, Wann, & Brown, 2006; Wilmut & Wann, 2008). Furthermore, Ayton et al. (2009), as well as Webber et al. (2011), did not find any link between the DEM test and saccadic parameters (accuracy, latency, speed), thus questioning the validity of the DEM test as a measure of oculomotor behavior during reading, but not as a measure of reading performance and speed of visual processing. Taking into account all these observations, some precautions are needed while using the DEM test as a measure of DCD's oculomotor behavior during reading. Alternatively, it is possible that oculomotor difficulties in DCD are

revealed depending on the task and the measures used. Thereby, it is worth to mention that studies reporting these atypicalities used different tasks and measures compared to what is asked for the execution of the DEM test. The former employed tasks such as pro- and anti-saccades, smooth pursuit, eye-hand coordination, walking, etc., and direct measures of saccadic parameters such as latency, speed, saccade targeting, number and duration of fixations and regressions, etc. Differently, as previously described, the DEM test is based on a RAN task and uses indirect measures such as errors and speed. Future studies should thus take into consideration the impact of the type of cognitive task on oculomotor control in DCD and develop more researches comparing indirect and direct measures of DCD children's eye-movements.

Limitations of the study

The present study unfortunately cannot disentangle the origin of the oculomotor difficulties in DCD between low-level processes or higher-level cognitive tasks (e.g., reading). As a matter of fact, our results showed that 1) performance at the DEM test are clearly associated to reading abilities, particularly to reading disorder and 2) the validity of the DEM test as a measure of oculomotor behavior is questionable. Consistently, it is also reasonable to observe that children with DCD made more errors in the DEM test because they have attentional difficulties which are frequently associated to this disorder (e.g., Dewey et al., 2002; Goulardins et al. 2017). Even if a more complete assessment of attentional functions should be necessary, we compared the scores at the ADHD questionnaire total score used for inclusion criteria between DCD, DCD+DD and DD groups. No differences emerged between DCD and other neurodevelopmental groups in terms of clinical attentional problems, suggesting that attentional problems could not explain DCD's performances at the DEM test. Surely, the question of

attention should be explored in more depth and deserves more consideration in future studies exploring eye-movements in DCD.

Finally, we relied on parental report, on a neuropsychological and on a medical assessment to check the visual functioning of the children in the present study. As children with neurodevelopmental disorders usually suffer from various debilitating visual impairments, it would be recommended to systematically test visual capability before testing visual perceptual skills.

Conclusions

Our study showed that 1) children with DCD presented mild atypical performances at the DEM test (error z-score only), 2) children with DD presented particularly poor performance on the DEM test, and 3) the co-morbid condition (DCD+DD) did not add to the severity of atypical performance obtained on the DEM test. In sum, children with DCD were the less affected according to the DEM test, and children with DD (isolated or comorbid) presented the most atypical performance. To go further, follow up research with direct measures of oculomotor control are required to 1) explore on-line oculomotor control during reading and/or visual word recognition in DCD (e.g., Bellocchi et al., 2015) and 2) measure the impact of DCD's oculomotor deficits on reading. In that sense, eye tracking should help researchers to explore the nature of oculomotor disorder in DCD children, distinguish DD and DCD oculomotor behavior and better apprehend DCD+DD performance variability.

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