

# 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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1	Special Issue	"VSI:DCD"

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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#### 26 Abstract

Studies have suggested a dysfunction in oculomotor skills in children with developmental 27 coordination disorder (DCD). It has been proposed that the Developmental Eye Movement 28 29 (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 30 31 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 32 specificity of the oculomotor difficulties, as measured by the DEM test. Results showed that 1) 33 children with DCD presented mild atypical performances at the DEM test (error z-score only), 2) 34 children with DD presented particularly poor performance on the DEM test, and 3) the co-morbid 35 condition (DCD+DD) did not add to the severity of atypical performance obtained on the DEM 36 test. In sum, children with DCD were the less affected according to the DEM test, and children 37 with DD (isolated or comorbid) presented the most atypical performance. Results at the DEM test 38 did not allow making emerge clear oculomotor atypicalities in DCD. We thus conclude that more 39 research using eye-tracking techniques is needed to explore the nature of oculomotor atypicalities 40 in DCD children, to distinguish DD and DCD oculomotor behavior, and to understand the profile 41 of children with dual diagnosis. 42

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

#### Introduction

Developmental Coordination Disorder (DCD) is a neurodevelopmental disorder 48 characterized by significant difficulties with the acquisition and execution of motor skill [DSM-5, 49 American Psychiatric Association (APA), 2013]. Individuals with DCD demonstrate a level of 50 motor skill out of keeping with their age and intellectual ability, in the absence of other medical 51 condition (Cerebral Palsy, sensory deficit, etc.). A meta-analysis showed a pattern of deficits 52 53 which are characteristic of DCD and involving different areas such as internal (forward) modelling, rhythmic coordination, executive functions, gait and postural control, catching and 54 interceptive action, and aspects of sensori-perceptual processing (Wilson, Ruddock, Smits-55 Engelsman, Polatajko, & Blank, 2013). Information processing has been shown to be altered in 56 this developmental disorder (Wilson and McKenzie, 1998). In particular, the authors identified 57 increased deficits with visuo-spatial processing tasks in the DCD population, regardless of 58 59 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, 60 2004; Biotteau, Albaret, Lelong, & Chaix, 2017; Gardner, 1982; Martin, 2006; Prunty, Barnett, 61 Wilmut, & Plumb, 2016). 62

Additionally, there is an increasing interest in literature over the last decades for eye-63 movement behavior in DCD while executing various tasks (Gaymard et al., 2017). Some studies 64 showed difficulties in pursuit tracking tasks in individuals with DCD, but the analysis of the 65 66 results also demonstrated preserved aspects (Langaas, Mon-Williams, Wann, Pascal, & 67 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) 68 reported a reduced gain in pursuit eye movements. More recently, Licari et al., (2018) found that 69 DCD individuals took longer to initiate smooth pursuit but once initiated, these individuals were 70

effectively able to maintain it. Furthermore, Robert et al. (2014) suggested a delay in the 71 72 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 73 only vertical pursuit gain was significantly lower in children with DCD. In addition to horizontal 74 75 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 76 horizontal pursuit gains were comparable in DCD and TD children. Moreover, response 77 preparation in the pro- and anti-saccade tasks were not impaired in children with DCD. The 78 authors concluded to intact fundamental neural mechanisms underlying pursuit and saccades in 79 DCD. However, children with DCD had deficits in maintaining engagement in the fixation and 80 pursuit tasks, and have problems with saccadic inhibition compared to TD controls of the same 81 age. Therefore, children with DCD would show oculomotor 'atypicalities' (Sumner et al.,  $2016)^1$ . 82 The eye-tracking technique has also been used to highlight difficulties in eye-hand 83 coordination in DCD. Langaas et al. (1998) showed that DCD children had difficulties in 84 smoothly synchronizing their eye movements with a moving object. Accordingly, Wilmut, Wann 85 and Brown (2006) observed that children with DCD have difficulties in concatenating the 86 sequential shifts of gaze and hand in order to complete everyday tasks or assessment items (i.e., 87 double-step pointing paradigm ; see also Katschmarsky, Cairney, Maruff, Wilson, & Currie, 88 2000, for similar results). Similarly, DCD children were found to have slower strategy of fixating 89 the target prior to initiating a hand movement (Wilmut & Wann, 2008). In the same vein, a 90

<sup>&</sup>lt;sup>1</sup> 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.

91 group-based gaze training intervention has shown to enhance DCD children's general

92 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

94 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' 97 eye-movements, the exploration of oculomotor control during reading in that specific population 98 is quite scarce in the literature. Nonetheless, reading difficulties have been observed in 29% to 99 70% of children diagnosed with DCD (O'Hare & Khalid, 2002). In this context, Bellocchi and 100 colleagues (2015) investigated the landing position pattern in oculomotor control that take place 101 in visual word perception in children with DCD. The authors asked children to perform an 102 103 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 104 middle of the stimulus and to validate this position by pressing a button. Even if the oculomotor 105 106 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, 107 Grainger, & Ducrot, 2019 for data on Developmental Dyslexia -DD<sup>2</sup>). Results showed that 108

<sup>&</sup>lt;sup>2</sup> *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 114 DCD may be explained or influenced by the presence of a co-occurring neurodevelopmental 115 disorder (see Smits-Engelsman, Jover, Green, Ferguson, & Wilson, 2017). Indeed, co-occurrence 116 between neurodevelopmental disorders is a very common condition. Because DCD often co-117 occurs with DD, eye movement disorders in this population could rely on reading subclinical 118 deficits. Indeed, epidemiological studies demonstrate a rate of comorbid diagnosis of DCD and 119 DD in 16% (Kaplan, Dewey, Crawford, & Wilson, 2001) to 70% of DCD children (Iversen, 120 Berg, Ellertsen, & Tønnessen, 2005). Similarly, Chaix and colleagues (2007) found an high 121 percentage of DCD diagnosis in a group of 58 dyslexics, i.e. 40% scored below -2 standard 122 deviations (SDs) on the Lincoln-Oseretsky Motor Development Scale (Rogé, 1984), and 17.2% 123 scored between -1 and -2 SDs. 124

Unfortunately, despite the evidence of a frequent association between DCD and DD, this 125 co-occurrence has not been taken into consideration in the research exploring oculomotor control 126 in these two clinical populations (i.e., DCD and DD). Bellocchi and colleagues (2015) did not 127 observe any difference between "DCD+DD" and "isolated DCD" children in the oculomotor 128 129 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 130 question of oculomotor control, recent studies exploring the impact of the co-occurrence between 131 DCD and DD on visuo-spatial abilities, have pointed out the absence of significant differences 132

133	between DD+DCD group and DCD or DD isolated groups in the Motor Reduced Visual
134	Perception and Visual-Motor Integration quotient components of the DTVP-2 (Bellocchi et al.,
135	2017), but also in visuo-attentional processes involved in word recognition (i.e., Optimal Viewing
136	Position <sup>3</sup> - OVP- Bellocchi & Ducrot, submitted). As a whole, these data suggest that the co-
137	morbid condition does not sharpen difficulties in eye movements or in visuo-spatial processing in
138	DCD children. Previous studies have reached the same conclusion, with no cumulative impact on
139	cognitive abilities in the case of a dual diagnosis (see Biotteau et al., 2017, for a detailed review;
140	Kaplan, Crawford, Cantell, Kooistra, & Dewey, 2006). However, recent findings depict a more
141	complex framework than what it looks like. Indeed, Bellocchi and Ducrot (under review)
142	suggested that children with dual diagnosis show an intermediate profile between children with
143	isolated disorders [see also Maziero, Tallet, Bellocchi, Jover, Chaix & Jucla (2020), for similar
144	results on working memory capacity]. It is clear that this point deserves a particular attention in
145	scientific studies. We will thus try to address that question here.

147 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

<sup>&</sup>lt;sup>3</sup> 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 153 154 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 155 abilities involved in a rapid automatized naming (RAN) task. It comprises horizontal and vertical 156 157 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 158 practice guideline (e.g., Facchin, Maffioletti, & Carnevali, 2011; Facchin, Ruffino, Facoetti, & 159 Maffioletti, 2014; Medland, Walter, & Woodhouse, 2010; Tassinari & DeLand, 2005). Another 160 advantage is that the DEM test provides four behavior profiles, which can disentangle RAN 161 difficulties from oculomotor dysfunction: 1) Type 1, which represents normal automaticity of 162 digit naming and oculomotor skills. 2) Type 2, which represents oculomotor dysfunction. 3) Type 163 3, which represents difficulties in automaticity of digit naming. 4) Type 4, which is a 164 165 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 166 factorial analysis showed saturation to three main factors. The first factor was linked to the 167 common naming component between vertical and adjusted horizontal time. This factor has been 168 thus referred to automatized naming skills. The second factor was related to adjusted horizontal 169 time and Ratio (i.e. the quotient between horizontal time and vertical time), and it has been 170 referred to oculomotor skills. They finally found a third factor constituted by Errors. These three 171 factors account for 99.8% of the total variance of the DEM test data. The authors concluded that 172 173 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 174 175 in naming and oculomotor skills, respectively.

176	Despite being criticized (e.g., Ayton, Abel, Fricke, & McBrien, 2009; Webber, Wood,
177	Gole, & Brown, 2011), the DEM test has recently been suggested to be useful in exploring the
178	oculomotor behavior of children with DD during reading. Moiroud, Gerard, Peyre, and Bucci
179	(2018) sought to associate the DEM test outcomes with measures of gaze behavior and showed
180	that children with DD and normal readers of equivalent reading level have significant longer
181	fixation times and take longer to read the horizontal reading task than normal readers of similar
182	chronological age. In addition, they found a correlation between the fixation time and the number
183	of words read in one minute with the total time of the horizontal reading task.
184	As a first goal, our work aimed at exploring oculomotor behavior in children with DCD as
185	assessed with the DEM test. Particularly, according to the interpretation of the results provided
186	by the DEM test, we predicted that if children with isolated DCD showed oculomotor difficulties,
187	they should mainly exhibit Type 2 profile that represents oculomotor dysfunction. In addition,
188	according to the so-called <i>phonological hypothesis</i> <sup>4</sup> (e.g., Snowling, 2000; 2006; Vellutino &
189	Fletcher, 2005; Vellutino, Fletcher, Snowling, & Scanlon, 2004), children with isolated DD
190	should be mostly linked to Type 3 profile which represents specific difficulties in RAN.
191	Subsequently, children with comorbid disorders (DCD+DD) should be mainly characterized by
192	Type 4 profile, which represents a combination of RAN and oculomotor skills difficulties.

<sup>&</sup>lt;sup>4</sup> 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.

193 Secondly, we wanted to explore the impact of comorbidity on DCD's oculomotor 194 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. 195 Pitcher, Piek, & Barrett, 2002), if co-morbid condition adds to the severity of the cognitive 196 197 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 198 199 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; 200 Biotteau et al., 2017). Moreover, the comparison between co-morbid children (DD+DCD) and 201 children with isolated disorders (DD or DCD) can provide evidences about the specificity of the 202 oculomotor difficulties, as measured by the DEM test, to reading or motor impairments. In other 203 words, if atypicalities at the DEM test are associated with reading deficit, we should find them 204 205 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 206 specifically in DCD children (isolated DCD and DD+DCD). 207 208 209

### Method

**Participants.** In total, 138 children (57 girls, 81 boys) participated in the study. Four 210 groups were constituted (see Table 1), one composed of 42 typically developing children (TD), 211 the other composed of 22 children with developmental coordination disorder (DCD), the third 212 213 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 214 215 patients studied were enrolled in the DYSTAC MAP cohort (ANR-13-APPR-0010).

216	All ch	ildren were a	nged between	7 years 8 mor	nths to 12 year	s 6 months	s, right-handed, and
217	French native	e speakers. A	ll parents repo	orted a norma	l hearing, and	normal or	corrected-to-normal
218	vision of thei	r child. All u	nderwent a fu	ll medical and	d neuropsychol	logical ass	essment conducted
219	by skilled pra	actitioners. T	hey all had no	ormal intellect	ual functioning	g level (sta	ndard score at least
220	above 7 at the	e Similarities	and Pictures	Concepts sub	test, French-la	nguage ve	rsion of the WISC-
221	IV, Wechsler	, 2005). Furt	hermore, child	dren whose of	al language sk	ills were in	n the pathological
222	range (scores	below -2 SE	on the « EV.	AC » -Flessas	& Lussier, 20	03- and «1	Ecosse » -Lecocq,
223	1996- tests),	or who were	diagnosed wi	th ADHD (siz	t or more symp	otoms of th	ne
224	« Hyperactivi	ity/impulsive	ness » as « In	attention » ch	ecklist of the I	OSM-5) w	ere excluded from
225	the study.				.0.	5.	
226					0		
227							
228							
229			2	[Insert Table	1 about here]		
			$\mathcal{O}_{\mathcal{N}}$			Group	
Grouj	þ	TD	DCD	DD	DCD+DD	diff	Post hoc comparison
N (fen	nales)	42 (20)	22 (7)	47 (20)	27 (10)	ns	
Age (I	M ±SD)	10,0 ±1,1	9,8 ±1,3	$10,1 \pm 1,1$	10,1 ±1,3	ns	
DSM- que tota ±S	5 ADHD estionnaire al score (M D)	0.86 ±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 ±3,1	20,84 ±4,8	5,45 ±2,7	20,33 ±5,3	<.001	TD;DD>DCD;DCD+DD
Alouette Accuracy Z-score (M ±SD)	0,57 ±0,3	0,32 ±0,4	-2,19 ±1,2	-2,24 ±1,9	<.001	TD>DCD>DD;DCD+DD

Alouette Speed Z- score (M ±SD)	0,95 ±0,8	0,31 ±0,8	-1,43 ±0,6	-1,35 ±0,5	<.001	TD>DCD>DD;DCD+DD
Odedys Irregular word Accuracy (M ±SD)	0,84 ±0,6	0,63 ±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
230						

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; DD= children with isolated DD; DCD+DD= children with both DCD and DD; Group diff= Group difference. Group differences were tested with a Kruskall Wallis test and  $\chi^2$ (sex ratio), and post-hoc test with Dwass-Steel-Critchlow-Fligner test.

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Participants with diagnosed neurodevelopmental disorders were recruited at San Salvator 239 University Hospital [Center for the diagnosis of learning disabilities] in Marseille, at the Hospital 240 of Aix-en-Provence, and at the Purpan University Hospital in Toulouse, France. Children with 241 DCD were included in the study if they had a score below the 5<sup>th</sup> percentile on the French version 242 of the M-ABC (Soppelsa & Albaret, 2004). Children with DD were included in the study if they 243 showed manifested reading deficits in two different tests : at least 1 SDs below the normal level 244 245 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 246

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 249 reading difficulties [at least -0.5 SDs above the normal level on the "Alouette Test-R" (Lefavrais, 250 251 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 252 motor difficulties [scores above the 15<sup>th</sup> percentile on the M-ABC test]. Children of the group 253 DCD+DD had a score below the 5<sup>th</sup> percentile on the M-ABC and reading impairments [i.e., 254 reading performance respecting the two previously presented criteria - at least 1 SDs below the 255 normal level on the "Alouette Test-R" and at least 1.5 SDs below the normal level on the 256 ODEDYS-2 test]. 257

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 15<sup>th</sup> 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.

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269 Materials. Each participant was given the DEM test (Garzia, et al., 1990) which is
270 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 271 272 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; 273 sixteen lines of five irregularly separated 0.5 cm high digits). Children were asked to read aloud 274 275 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) 276 277 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) 278 the adjusted horizontal time (second) (HT) which represents the time required for reading the 279 eighty horizontally organized digits presented in the Test C; this score is corrected for omission 280 or addition errors<sup>5</sup>; 3) the total number of errors which gives the accuracy on the execution of 281 Test C (errors recorded : omission, addition, substitution, transposition); 4) the Ratio score which 282 is calculated dividing the HT by the VT. 283

Furthermore, for clinical purposes, the DEM test provides four behavior profiles: 1) Type 284 1, which represent normal automaticity of digit naming and oculomotor skills. It comprises 285 essentially normal performance in HT, VT and Ratio. The children were included in this group if 286 all the scores were > -1.5 SD. 2) Type 2, which represents oculomotor dysfunction. It is 287 characterized by abnormally increased scores in HT and Ratio but normal scores for VT. The 288 children were included in this group if HT and Ratio scores were  $\leq -1.5$  SD and VT scores > -289 1.5 SD. 3) Type 3, which represents difficulties in automaticity of digit naming. It comprises 290 291 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  $\leq -1.5$  SD and Ratio scores > -1.5 SD. 4) Type 292

<sup>&</sup>lt;sup>5</sup> Adjusted Horizontal Time = Test C time x [80/(80-number of omission + number of addition)]

293	4, is a combination of Type 2 and 3 and represents difficulties in digit naming and oculomotor
294	skills. It is characterized by abnormally increased scores in VT, HT and Ratio. The children were
295	included in this group if all the scores were $\leq -1.5$ SD.
296	
297	Procedure. All children were tested individually at the hospital and at the university
298	laboratories by a neuropsychologist and a psychomotor therapist enrolled in the research project
299	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.

302

300

Data Analysis. Data were analysed using the Jamovi statistical computer software [The 303 304 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 305 306 were small, all statistics were non-parametric. Group comparisons for the children's test scores 307 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 308 comparisons were done with Dwass-Steel-Critchlow-Fligner test which compares all pairs of 309 groups using a pairwise ranking nonparametric method and controls the error rate. The 310 311 distribution of the sex ratio and of the DEM types between the groups were tested using  $\chi_2$  tests 312 and pairwise comparisons were performed with Bonferroni correction (p = .008). The significance threshold was set at p = .05. 313

314

315 **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.

323



[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.

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The distribution of the behavior types differed significantly between the groups of children ( $\chi_2 = 37.5$ , 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.

- 338
- 339



[Insert Figure 2 about here]

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Figure 2: Distribution of the four behavior types in each group. Note: 1) Type 1: normal 342 automaticity and oculomotor skills. 2) Type 2: oculomotor dysfunction. 3) Type 3: difficulties in 343 automaticity of digit naming. 4) Type 4: difficulties in digit naming and oculomotor skills (a 344 combination of Type 2 and 3). TD = typically developing children; DCD= children with isolated 345 346 DCD; DD= children with isolated DD; DCD+DD= children with both DCD and DD. 347 Discussion 348 The first aim of the present study was to explore oculomotor behavior in children with 349 DCD as assessed with the DEM test (Garzia et al., 1990). As a whole, results showed that 350 children with DCD presented slight atypical scores at the DEM test, as attested by the fact that 351 only one parameter was significantly affected. Particularly, children with isolated DCD made 352 more errors if compared to TD children. Children with DCD+DD clearly showed a wider range 353

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)<sup>6</sup>. 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

<sup>&</sup>lt;sup>6</sup> 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.

Contrary to our expectations, the results of the present study showed that atypical scores 364 365 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 366 children with isolated DD, showed a wider range of difficulties if compared to TD children. 367 Indeed, they not only made more errors than the DCD only group, but also showed lower scores 368 for the horizontal time, vertical time, ratio and atypical oculomotor responses if compared to TD 369 children (i.e., higher presence of Type 2 and 4). In other words, the results at the DEM test were 370 very sensitive to children's reading abilities or, more specifically, to the presence of a reading 371 disorder. This is what was found by Moiroud and collaborators (2018) who showed that dyslexic 372 373 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 374 and reading age-matched children had longer fixation time compared to chronological age-375 376 matched children while reading this text. Accordingly, previous important findings showed that compared to age-matched control readers, dyslexics' eye movements in word, pseudoword, or 377 sentence reading are characterized by more and longer fixations. Additionally, dyslexics usually 378 show shorter saccades and more regressions (Bellocchi et al., 2019; Biscaldi, Gezeck, & Stuhr, 379 1998; Bucci, Nassib, Gerard, Bui-Quoc, & Seassau, 2012; Hawelka, Gagl, & Wimmer, 2010; 380 381 Hutzler & Wimmer, 2004; McConkie, Zola, Grimes, Kerr, Bryant & Wolff, 1991; Rayner, 1986; Seassau, Gerard, Bui-Quoc, & Bucci, 2014). 382

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.

Until now very few studies have investigated whether the oculomotor control atypicalities 392 found in DCD may be, or not, influenced by the presence of a co-occurring DD. Considering the 393 evidence of a frequent association between these two neurodevelopmental disorders (Iversen et 394 al., 2005; Kaplan et al., 2001), our second aim here was to explore the impact of comorbidity of 395 DD on DCD's oculomotor skills, particularly the oculomotor skills measured by the DEM test. 396 As a whole, results showed no significant differences between comorbid and isolated groups at 397 the DEM test. This means that no additional impact on DEM performance is associated with a 398 dual diagnosis. Therefore, these data do not support the cumulative hypothesis according to 399 400 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 401 (e.g., Pitcher et al., 2002). These results are in line with other recent studies suggesting that the 402 403 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., 404 405 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 406 disorders. Accordingly to what it has been stated above, this result can be mainly explained by 407 the presence of a co-occurred DD. 408

409	Even if the differences between the three groups of children with neurodevelopmental
410	disorders (DCD, DCD+DD, and DD) were not statistically significant, it does not mean that these
411	groups had comparable performance on the DEM test. Indeed, if we look at the distribution of the
412	behavior types (Figure 2), it appears that the profiles that emerged are quite heterogeneous.
413	Particularly, we can observe that DCD+DD group showed higher percentage of Type 4 profile
414	compared to the other two profiles (Type 2 and 3) and compared to isolated DD and DCD. Again,
415	it is important to notice that these differences were not statistically significant but they highlight a
416	variability that researchers need to explore more deeply as it could depict a more complex
417	framework of the DCD+DD comorbid profile.
418	Finally, results at the DEM test did not reveal clear oculomotor atypicalities in DCD. For
419	instance, the percentage of Type 2 profile, indicating an oculomotor dysfunction, wasn't higher in
420	children presenting a DCD isolated or comorbid, compared to the other neurodevelopmental
421	groups. Rather than concluding that DCD children do not encounter eye-movements difficulties,
422	we assume that the DEM test is not as sensitive to oculomotor than to RAN difficulties. Indeed,
423	first of all, the vast majority of researches observed oculomotor difficulties in DCD (e.g.,
424	Bellocchi et al., 2015; Gaymard et al., 2017; Gonzalez et al., 2016; Langaas et al., 1998;
425	Katschmarsky et al., 2000; Licari et al., 2018; Lord & Hulme, 1988; Robert et al., 2014; Sumner,
426	et al. 2016; Warlop et al., 2020 ; Wilmut, Wann, & Brown, 2006; Wilmut & Wann, 2008).
427	Furthermore, Ayton et al. (2009), as well as Webber et al. (2011), did not find any link between
428	the DEM test and saccadic parameters (accuracy, latency, speed), thus questioning the validity of
429	the DEM test as a measure of oculomotor behavior during reading, but not as a measure of
430	reading performance and speed of visual processing. Taking into account all these observations,
431	some precautions are needed while using the DEM test as a measure of DCD's oculomotor
432	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 433 434 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, 435 smooth pursuit, eve-hand coordination, walking, etc., and direct measures of saccadic parameters 436 437 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 438 439 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 440 comparing indirect and direct measures of DCD children's eye-movements. 441

442

443 Limitations of the study

The present study unfortunately cannot disentangle the origin of the oculomotor 444 445 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 446 to reading abilities, particularly to reading disorder and 2) the validity of the DEM test as a 447 measure of oculomotor behavior is questionable. Consistently, it is also reasonable to observe 448 that children with DCD made more errors in the DEM test because they have attentional 449 difficulties which are frequently associated to this disorder (e.g., Dewey et al., 2002; Goulardins 450 et al. 2017). Even if a more complete assessment of attentional functions should be necessary, we 451 compared the scores at the ADHD questionnaire total score used for inclusion criteria between 452 453 DCD, DCD+DD and DD groups. No differences emerged between DCD and other neurodevelopmental groups in terms of clinical attentional problems, suggesting that attentional 454 problems could not explain DCD's performances at the DEM test. Surely, the question of 455

456 attention should be explored in more depth and deserves more consideration in future studies 457 exploring eye-movements in DCD.

Finally, we relied on parental report, on a neuropsychological and on a medical 458 assessment to check the visual functioning of the children in the present study. As children with 459 460 neurodevelopmental disorders usually suffer from various debilitating visual impairments, it would be recommended to systematically test visual capability before testing visual perceptual 461 skills. 462

463

#### Conclusions 464

Our study showed that 1) children with DCD presented mild atypical performances at the 465 DEM test (error z-score only), 2) children with DD presented particularly poor performance on 466 the DEM test, and 3) the co-morbid condition (DCD+DD) did not add to the severity of atypical 467 performance obtained on the DEM test. In sum, children with DCD were the less affected 468 according to the DEM test, and children with DD (isolated or comorbid) presented the most 469 atypical performance. To go further, follow up research with direct measures of oculomotor 470 control are required to 1) explore on-line oculomotor control during reading and/or visual word 471 recognition in DCD (e.g., Bellocchi et al., 2015) and 2) measure the impact of DCD's oculomotor 472 deficits on reading. In that sense, eye tracking should help researchers to explore the nature of 473 474 oculomotor disorder in DCD children, distinguish DD and DCD oculomotor behavior and better apprehend DCD+DD performance variability. 475

476

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