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Dysgraphia Differs Between Children with Developmental Coordination Disorder

and/or Reading Disorder

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CONFLICTS OF INTERESTS

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Abstract

Handwriting deficits, or dysgraphia, are present in several neurodevelopmental disorders. To investigate whether dysgraphia differs according to the associated disorder, we performed a detailed analysis of handwriting in children with developmental coordination disorders (DCD), reading disorder (RD), or comorbid RD and DCD. Handwriting deficits were investigated at the product (quality of the trace) and the process (movement that generates the trace) levels. Nineteen children with singular RD (among which 8 with dysgraphia), 13 children with singular DCD (among which 7 with dysgraphia), 16 children with comorbid RD+DCD (among which 11 with dysgraphia), and 20 typically developing children, age 7 to 12, performed the BHK (Brave Handwriting Kinder) test, a standardized assessment of handwriting, on a graphic tablet. DCD primarily affected handwriting quality, while RD affected slowness and, to a lesser extent, quality. Children with RD, solely or comorbid with DCD, wasted time by lifting and stopping the pen when writing. The comorbidity added to but did not worsen, handwriting difficulties. These results reflect distinct motor impairments and/or strategies in children with DCD or RD. We identified subtypes of dysgraphia and advocated for a fine-grained analysis of the writing process and the assessment of motor and reading skills when studying dysgraphia.

Keywords: DCD, dyslexia, dysgraphia, handwriting deficits, fine-grained analysis, reading disorder

Abbreviations: DCD: developmental coordination disorders; RD: reading disorder; DG: dysgraphia; CTL: control; BHK: Brave Handwriting Kinder; SNvpd: Signal-to-Noise velocity peaks difference.

Dysgraphia Differs Between Children with Developmental Coordination Disorder and/or Reading Disorder

Handwriting deficits, characterized by poor quality and/or speed which cannot be explained by a neurological or intelligence deficit, or a lack of writing practice, are referred to as dysgraphia. If not identified and handled early, these deficits can have a serious impact on children's academic performance. Dysgraphia does not constitute a discrete disorder according to international classifications (American Psychiatric Association, 2013) but is reported in several neurodevelopmental disorders. The first question addressed in this study is, whether the nature of dysgraphia differs between Developmental Coordination Disorders (DCD) and Reading Disorder (RD), both of which are neurodevelopmental disorders involving distinct, if not independent, impairments.

The comorbidity between neurodevelopmental disorders, such as RD, Attention Deficit Hyperactivity Disorder (ADHD), or DCD is often observed (Kaplan et al., 2001; Ramus et al., 2003; Rochelle et al., 2009). Whilst DCD and RD concern distinct domains (reading vs motor coordination), they often appear in a comorbid manner (Pennington et al., 2019; Smits-Engelsman et al., 2017). The processes underlying this heterotopic comorbidity are still poorly documented and understood (Dewey, 2018). Furthermore, the impact of this comorbid condition on the children's potential handwriting deficits is unknown and constitutes the second question addressed in this study.

Dysgraphia in Children with DCD

The association between dysgraphia and DCD has long been observed (for a recent review, see Barnett & Prunty, 2020). Depending on the study, handwriting deficits have been found in 50%-88% of children with DCD (Di Brina et al., 2021; Huau et al., 2015; Lopez et al., 2018; O'Hare & Khalid, 2002; Vaivre-Douret et al., 2011). Handwriting difficulties also

constitute the second reason for medical consultation (Geuze, 2005; Miller et al., 2001). Many studies have documented handwriting deficits in children with DCD, both at the product level, *i.e.*, the quality of the written trace, and at the process level, *i.e.*, the movement that generates the trace. At the product level, a decreased legibility, an increased number of corrections, and a random spatial arrangement of the letters demonstrate a lower handwriting quality in children with DCD compared to control (CTL) children (Di Brina et al., 2021; Rosenblum & Livneh-Zirinski, 2008). The letters are of irregular shape, often too large, and less consistent than those of CTL children (Bo et al., 2014; Di Brina et al., 2021; Huau et al., 2015; Jolly & Gentaz; 2014; Prunty & Barnett, 2020). At the process level, children with DCD make more pauses during writing, both 'on paper' (more stops) and 'in-air' (more lifts; Prunty et al., 2013, 2014; Rosenblum et al., 2003; Smits-Engelsman et al., 2001). Likewise, on-paper and lift durations were longer in children with DCD in a copy task (Rosenblum & Livneh-Zirinski, 2008; Rosenblum & Regev, 2013). Finally, they also fail to adapt to size or speed constraints (Huau et al., 2015; Smits-Engelsman et al., 2001).

The examination of handwriting in DCD children however led to some discrepancies. Pen pressure on the writing surface is either lower or higher in children with DCD compared to children without DCD (Di Brina et al., 2008; Prunty et al., 2020; Rosenblum & Livneh-Zirinski, 2008). Furthermore, some authors found a higher writing speed in children with DCD (Jolly & Gentaz, 2014) while others did not (Prunty et al., 2013), or found a lower speed (Di Brina et al., 2021).

Dysgraphia in DCD children has mainly been explained by a lack of movement sequence automation and motor planning in motor skills (Barnett & Prunty, 2020; Huau et al., 2015; Prunty & Barnett, 2020) and/or a neuromotor noise and a deficit in predictive motor control (for a review, see Adams et al., 2014). More precisely, the variability in handwriting

performance of children with DCD suggests a lack of exploiting the noise-reducing capabilities of muscles and joints (van Galen et al., 1993).

Dysgraphia in Children with RD

Handwriting deficits in children with RD have been less investigated, and more discrepancies remain in the literature (for reviews see Döhla & Heim, 2016; Jover et al., 2013). At the product level, children with RD write larger and with poorer legibility than CTL children, due to more missing and/or concatenated strokes within letters (Alamargot et al., 2020; Capellini et al., 2010; Lam et al., 2011; Sovik et al., 1987). At the process level, some studies reported lower writing speed and pen pressure (Afonso et al., 2020; Cheng-Lai et al., 2013; Pagliarini et al., 2015), increased lift duration (Brun-Hénin et al., 2013), increased total writing time, and a higher number of pauses (Alamargot et al., 2020) in children with RD compared to children without RD. Likewise, results concerning writing speed in children with RD are not unequivocal, some authors show a lower speed in children with RD while others do not (Afonso et al., 2020; Pagliarini et al., 2015; Sumner et al., 2013).

To what extent do these handwriting difficulties in children with RD result from slower language/orthographic processing (Afonso et al., 2020; Arfé et al., 2020; Sumner et al., 2014), or from graphomotor deficits in addition to these processing difficulties at a more central level (Gosse et al., 2022; Downing & Caravolas, 2023; Jover & Huau 2021), is still debated. As motor skills were not always checked in children with RD, it is still unclear whether their handwriting difficulties arise from their spelling deficit or motor difficulties *per se* (Gosse & Van Reybroeck, 2020). For instance, atypical pausing behaviors amongst children with RD were associated with a lack of automaticity in spelling (Prunty et al., 2013; Sumner et al., 2013, 2014) but could also arise from an associated undetected DCD.

Dysgraphia in Children with RD+DCD

Comorbidity between neurodevelopmental disorders is very frequent and constitutes a line of research to better understand the developmental trajectory of the deficits and their underlying mechanisms (Dewey, 2018). Studies systematically exploring the effect of comorbidity are recent (Bellocchi et al., 2021; Biotteau et al., 2017a, 2017b; Cignetti et al., 2018; Downing & Caravolas, 2020; Maziero et al., 2020). The prevalence of comorbidity between RD and DCD is higher than the chance level but is not yet precisely known. Recent studies on different groups of children showed that for most tasks, children with comorbid DCD and RD did not differ from children presenting with isolated RD or DCD: working memory (Maziero et al., 2020), oculomotor behavior (Bellocchi et al., 2017, 2021; Bellocchi & Ducrot, 2021), graphomotor tasks (Jover & Huau, 2021). One exception should, however, be mentioned: anticipatory postural adjustments were worsened in children of the comorbid group as compared to the RD- or DCD-alone groups (Cignetti et al., 2018). These results highlight an additive effect, which was also detected in Downing's (2018) study, and are in line with the multiple deficits approach to neurodevelopmental disorders (Pennington, 2006; Pennington et al., 2019).

The effect of comorbidity between RD and DCD on children's handwriting has been barely explored. Cheng-Lai et al. (2013) showed that RD children with a comorbid condition (DCD or ADHD) have a reduced handwriting speed compared to children with singular RD. Downing specifically explored the effect of comorbidity on RD and/or DCD children's handwriting (Downing, 2018; Downing & Caravolas, 2018). The authors showed that children with RD had lower scores on some dimensions related to handwriting legibility (letter formation and word spacing). In contrast, children with DCD had lower scores on all dimensions of handwriting legibility. The handwriting of children with comorbid DCD and RD did not differ from that of children with singular disorders, suggesting that their difficulties reflect an additive profile of spelling and additional, DCD-related, deficits. Using

a dimensional approach and generalized additive model analysis, Jover and Huau (2021) analyzed the handwriting of children with or without DCD and RD. They showed that handwriting difficulties depended on the children's scores on both motor and reading tests. Comorbidity added but not multiplicated handwriting difficulties.

Aim and Hypothesis

There are few studies comparing handwriting process and product deficits in children with RD or DCD children in a common protocol. The assessment of handwriting is often based on paper-and-pencil tests, which prevents a detailed analysis of the variables characterizing the process of handwriting (Danna et al., 2013). In addition, the question of the impact of comorbidity on handwriting performance has seldom been addressed. Identifying and understanding the deficits specific to each disorder is important from a clinical perspective, both to enable more efficient and well-targeted remediation and to help in the diagnosis of dysgraphia (Asselborn et al., 2018; Gargot et al., 2020).

This study aimed to assess whether dysgraphia differs according to the associated neurodevelopmental disorder, based on a digitalized analysis of the BHK test (Hamstra-Bletz et al., 1987; French version by Charles et al., 2003). The BHK test produces two scores, namely a quality score based on 13 spatial criteria related to the written trace, and a speed score based on one single criterion measuring the number of characters (letters and punctuations) produced in five minutes. A more detailed analysis of the process of writing in the spatial, temporal, kinematic, and dynamic aspects is required to complement this evaluation mainly based on the legibility of the written trace, and to better identify differences related to the neurodevelopmental disorder. First, the quality and speed scores of the BHK test were rated to determine the handwriting performance and the proportion of dysgraphia in children with DCD and/or RD and CTL children. Then, we compared children with

dysgraphia and RD, DCD, or RD+DCD to identify subtypes of dysgraphia. We hypothesized that children with DCD and children with RD would encounter different difficulties reflected by distinct handwriting features. More specifically, while the reduction in writing speed in children with RD is relatively well established in the literature, the results are more controversial with regards to DCD. More fine-grained analysis of the writing movement recorded on the tablet will enable us to determine precisely what changes in the process of forming the written trace are relative to DCD and RD. The question is to determine whether the writing difficulties related to each disorder are qualitatively different in terms of pen lifts and stops, movement fluency, and trace length. Furthermore, we expected that comorbidity would result in the addition of RD and DCD-specific difficulties but would not worsen the difficulties associated with each disorder.

Method

Participants

Sixty-eight children were included in the study, divided into four groups: the 'RD' group of 19 children with singular reading disorder, the 'DCD' group of 13 children with singular developmental coordination disorder, the 'RD+DCD' group of 16 children with comorbid RD and DCD, and the 'CTL' group of 20 children without reading or motor difficulties. Children with RD and/or DCD had received their diagnosis beforehand from an experienced clinician based on the DSM5 criteria (APA, 2013) and were receiving treatment for a neurodevelopmental disorder. Demographic and clinical profiles for the groups are available in Table 1. All children were right-handed and French native speakers. There was no difference in mean age ($\chi^2 = 2.84$, df = 3, p = .417) nor in gender proportion ($\chi^2 = 1.25$, df = 3, p = .74) between the groups. All parents reported normal hearing and normal or corrected-to-normal vision.

INSERT TABLE 1 HERE

All children underwent complete medical and psychological screening before their inclusion in the study (see Table 1). We used the French version of the WISC-IV (Wechsler, 2005) to establish their normal intellectual functioning levels. We referred to the Full-Scale IQ, when available, or the Similarities and Pictures Concepts subtests. These subtests belong to the Verbal Comprehension Index and to the Fluid Reasoning Index respectively. In the French version they have demonstrated good reliability (.77 and .64) and acceptable convergent validity (.58 and .50 respectively). They are considered complementary measures of general intelligence (Grégoire et al., 2006; Keith et al., 2006). The subtests' raw scores were converted to age-scaled standard scores (M = 10, SD = 3) and children were included if their scores were equal or above 7. Although all children reached this score, it is worth noting that there was a significant difference in the Similarities subtest score of the WISC between the CTL group and the two groups with RD. This difference is not surprising due to the nature of the Similarities task which relies strongly on lexicon knowledge (Wechsler, 2005). Preserved oral language skills were established using two tests. The subtest 'missing words' of the EVAC (Epreuve Verbale d'Aptitude Cognitive; Flessas & Lussier, 2003) evaluates the morpho-syntactic abilities on the expressive side and requires the children to write a missing word in a sentence (e.g., Il est recommandé de se brosser les dents d'aller dormir / It is recommended to brush your teeth ____ going to bed). The test was standardized on 943 children between 7 and 16 years but its psychometric properties haven't been further tested. On the other hand, the ECOSSE (Epreuve de Compréhension Syntaxico-Sémantique; Lecocq, 1996), based on Bishop's (1983) test of Reception of Grammar, evaluates the syntacticsemantic comprehension. It requires the child to point to a picture corresponding to a word or a sentence pronounced by the experimenter (e.g., L'homme poursuit le chien / The man chases the dog). The test was standardized on 2000 children between 7 and 12 years, however its

psychometric properties haven't been tested. Finally, attention-deficit / hyperactivity disorder (ADHD) was excluded on the basis of the parent's responses to the DSM-5 checklist (American Psychiatric Association, 2013).

The children were also evaluated regarding their reading and motor skills. The Alouette test (Lefavrais, 2005) was used to evaluate reading accuracy and efficiency (accuracy/reading time). The text to read aloud in this test has no meaning and requires word decoding. The Alouette is the most commonly used standardized test for the evaluation of reading capacity in France. It was standardized on 415 children between 6 and 16 years, but its psychometric properties haven't been tested.

Reading and phonological abilities were evaluated with items of the first and second versions of the ODEDYS test (Jacquier-Roux et al., 2005). Reading abilities were tested with logatom reading (reading isolated pseudo words) and word reading (reading isolated irregular words). Both reading accuracy and speed were scored. Phonological abilities were tested using 3 items of the version 2 of ODEDYS: phoneme suppression (remove the first phoneme of a word to repeat), phoneme fusion (build a pseudo word using the first phoneme of two words), and logatom repeating (repeat pseudo words after the experimenter). The ODEDYS test was standardized on 536 children between the 2nd and the 5th grades but the psychometric properties haven't been tested.

Motor skills were assessed using the Movement Assessment Battery for Children 1st Ed. (Soppelsa & Albaret, 2004). The French norms of the MABC were based on 1233 children between 4 and 12 years. The reliability was satisfactory (89% of agreement), the convergent validity was low, although significant (r = .40).

The following criteria were used to compose the groups. Children with RD (singular or associated with DCD) had received their diagnosis from an experienced clinician and were

receiving treatment for a reading problem by a pediatric speech therapist. They scored below the normal range when reading isolated irregular words or logatoms (-1.5 SD; ODEDYS) and/or when reading a meaningless text (-1 SD; Alouette test). Children with DCD (singular or associated with RD) had received their diagnosis from an experienced clinician and were receiving treatment for a motor coordination problem that interfered with their daily living activities and scored below the 15th percentile at the MABC1, which corresponds to the cut-off value for moderate DCD (for a review, see Smits-Engelsman et al., 2015). To avoid any overlap between groups, additional inclusion criteria included a MABC1 score above the 20th percentile for RD and CTL children and reading scores above -0.5 SD on reading tests for DCD and CTL children.

Children were enrolled in the cohort for a large multidimensional research project (DYSTAC-MAP cohort, ANR-13-APPR-0010). Children with RD and/or DCD were recruited via speech or psychomotor therapists with whom they were undergoing rehabilitation at local hospitals, or through public announcement. Children of the CTL group were recruited through public announcements. The parents and children gave their written informed consent to participate in the study before the start of the project, which had been approved by the French Ethics Committee Review Board (CPP, agreement 2014-A01960-47).

Task

Children took the test during their first visit to the laboratory, around one week after their inclusion visit. The children performed the BHK test (Hamstra-Bletz et al., 1987), a standardized evaluation of handwriting quality and speed. The task requires children to copy out a text for five minutes on a blank paper, with their usual handwriting. The handwriting quality score is calculated based on 13 criteria described in the manual, resulting in a degradation score: the higher the score, the worse the quality. These criteria are: 1. Writing is

too large; 2. Widening of left-hand margin; 3. Bad letter or word alignment; 4. Insufficient word spacing; 5. Acute turns in connecting joints to letters; 6. Irregularities or absence of joints (breaks in the trace); 7. Collisions of letters; 8. Inconsistent letter size; 9. Incorrect relative height of the various kinds of letters; 10. Letter distortions; 11. Ambiguous letter forms; 12. Corrections of letter forms; 13. Unsteady writing trace. A detailed description of each criterion can be found in the pioneer studies by Hamstra-Bletz and colleagues (Hamstra-Bletz et al., 1987; Hamstra-Bletz & Blöte, 1990). Only the five first lines of the text are rated. Each criterion is score between 0 and 5. The five lines are analysed together for criteria 1 and 2. Each line is analysed separately for criteria 3 to 13, with a score of 0 or 1 for each line depending if the criterion is absent or present in the line. The final quality score is the sum of all subscores. The handwriting speed score is estimated by counting the number of written characters. The French adaptation of the test demonstrated satisfactory concurrent validity (r = .68), intra- (percentage of agreement = 80 to 92%) and inter-rater reliability (r = .68-.9) (Charles et al., 2003). The BHK test was performed under the usual conditions, except that the children wrote the text on a sheet of paper (29.7 cm x 21 cm) affixed to a graphic tablet (Wacom®, Intuos 4L, 200 Hz). Digital data were recorded by a Matlab® program including the Psychophysics toolbox (http://psychtoolbox.org/). The BHK tests were rated by an experienced clinician (Charles et al., 2003). Reliability was assessed with a second rating performed by CJ on 25% of the sample. Intraclass Correlation Coefficient (ICC) was 0.85 for the quality score and 1.00 for the speed score, indicating very good reliability.

Data Analyses

Clinical analysis

The quality and speed *z*-scores were compared between the groups of children (CTL, RD, DCD, RD+DCD). Furthermore, based on these scores, we identified the children with

dysgraphia in each group. Children were considered with dysgraphia if their quality *z*-score was greater than or equal to +1.5 *SD* (deterioration score) or if their speed *z*-score was lower than or equal to -1.5 *SD* (Table 1). Note that none of the children in the CTL group reached those thresholds. We thus obtained three subgroups of children which all displayed dysgraphia: children with singular RD (DG-RD), children with singular DCD (DG-DCD), and those with comorbid RD and DCD (DG-RD+DCD).

Digital analysis

As only the first five lines of the produced text are considered for the clinical evaluation, we selected this part of the text for the digital analysis of handwriting using the recorded (x, y) coordinates, time, and pen pressure. Data segmentation of the written tracks was conducted with a JAVA program to dissociate tracings and pen lifts. The six digital variables extracted from the written tracks are presented in Table 2. These variables have been extensively investigated for examining the handwriting process in children with and without learning disabilities (for a brief review, see Danna et al., 2013). They respectively describe the spatial (trace length), temporal (tracing duration, total lift time, and total stop duration), kinematic (SNvpd) and dynamic (mean pen pressure) content of handwriting performance.

INSERT TABLE 2 HERE

Statistical Analyses

Data were analyzed using Jamovi (<u>https://www.jamovi.org</u>). Firstly, a non-parametric Kruskal-Wallis test with the four groups of children (CTL, RD, DCD, and RD+DCD) as between-subjects factor was conducted to compare the age and the BHK quality and speed *z*-scores. Dwass-Steel-Critchlow-Fligner tests were applied for post hoc pairwise comparisons. Effect sizes were calculated using epsilon squared (ε^2). Chi2 tests (χ^2) were used to compare

the proportion of males/females and the proportion of children with dysgraphia between groups. Secondly, we compared the three subgroups of children with dysgraphia with a Kruskal-Wallis test because of their reduced number, and Dwass-Steel-Critchlow-Fligner for post-hoc pairwise comparisons. The Benjamini-Hochberg method was used to adjust p values for multiple testing (q < 0.25). All significance levels were set at p = 0.031. Effect sizes were expressed using epsilon squared (ϵ^2).

Results

BHK z-scores

This first analysis aims at comparing the children's overall writing performance between the groups. Results are presented in Figure 1. Median and interquartile range values are presented in Table S1. A group effect was observed for both the quality and speed scores $(F(3,64)=12.1,\,p<.001,\,\eta^2_{\,p}=.363;\,F(3,64)=10.1;\,p<.001,\,\eta^2_{\,p}=.321$ respectively). Concerning the quality score, post-hoc comparisons revealed significant differences between the CTL group and the RD group (p = .013), and between CTL group and both the DCD and RD+DCD groups (p < .001), the latter displaying the worst scores (see Table S2 for complete results). Concerning the speed score, post hoc comparisons revealed differences between the CTL group and both the RD and RD+DCD groups (p < .001) (see Table S3 for complete results). Again, the RD+DCD group displayed the worst BHK speed scores.

INSERT FIGURE 1 HERE

Dysgraphia Proportion in Neurodevelopmental Disorders

Fifty-four percents of the children with neurodevelopmental disorders had BHK scores reaching one of the clinical thresholds for dysgraphia. The dysgraphic children were 8 out of 19 in the RD group (42.1%), 7 out of 13 in the DCD group (53.8%), and 11 out of 16 in the

RD+DCD group (68.7%). Only three children did not present deficits in their BHK quality score and were included in the group with dysgraphia solely on the basis of their velocity score. These three children all presented a RD. All other children presented deficits in the BHK quality score, or in both scores. No children from the CTL group reached one of the thresholds for dysgraphia. The proportion of children with dysgraphia did not differ significantly between the 3 groups of children with neurodevelopmental disorders ($\chi^2 = 2.483$, df = 2, p > .2).

Dysgraphia and Neurodevelopmental Disorders

To better understand the handwriting deficits depending on the neurodevelopmental disorder, we compared the clinical and digital variables between the subgroups of RD, DCD, and RD+DCD children with dysgraphia (DG-RD, DG-DCD, and DG-RD+DCD respectively). Results are presented in Table 3.

INSERT TABLE 3 HERE

Concerning the BHK scores, no significant difference between groups was observed for the quality or the speed score. Concerning the digital analysis, four kinematic features were significantly different between the subgroups of children with dysgraphia: the tracing duration, the total lift duration, the total stop duration, and the SNvpd. Post-hoc analyses (Table 4) showed that these four parameters were lower in DG-DCD children than in DG-RD, and that the total lift duration, the total stop duration and the SNvpd were lower in DG-DCD than in DG-RD+DCD children. Note that the DG-RD+DCD children differentiated themselves only from children with DG-DCD and that the former group of children showed a much higher interquartile range than the latter.

INSERT TABLE 4 HERE

Discussion

Identifying and understanding the underlying deficits of dysgraphia is important for both well-targeted diagnosis and more efficient rehabilitation. In this study, we performed a fine-grained analysis of handwriting in children with DCD, RD, or comorbid DCD and RD, both at the product and at the process levels, using the BHK test recorded with a graphic tablet.

Assessment of Handwriting among Neurodevelopmental Disorders

We found significant differences when comparing BHK scores among the groups of children. Children with DCD obtained the worst quality scores, while children with RD, either singular or combined with DCD, had the worst speed scores. In agreement with previous observations, children with singular RD wrote more slowly and with a relatively lower quality than the control children (Alamargot et al., 2020; Capellini et al., 2010; Lam et al., 2011; Sovik et al., 1987; Sumner et al., 2013). The nature of the BHK task, which involves reading, may partly explain the writing slowness of children with RD. In addition, we found that children with singular DCD presented a very poor handwriting quality as described earlier (Bo et al., 2014; Di Brina et al., 2021; Huau et al., 2015; Jolly & Gentaz; 2014; Prunty & Barnett, 2020; Rosenblum & Livneh-Zirinski, 2008). The DCD children did not write significantly fewer characters than the control children as the difference between CTL and DCD children was not significant for the BHK speed score. As mentioned in the introduction, discrepancies regarding writing speed were found among the publications in children with DCD (Di Brina et al., 2021; Jolly & Gentaz, 2014; Prunty et al., 2013). Our results are congruent with those of Prunty et al. (2013). Using the Italian adaptation of the BHK, Di Brina et al. (2021) showed that quality and speed scores were lower in children with DCD than in children without DCD. As this team did not control for comorbidity, one possible explanation may be the presence of

comorbid RD which may also impact handwriting in the Italian DCD group. Although they all presented noticeable difficulties in the handwriting assessment, our results showed that handwriting difficulties between children with DCD and children with RD differed. Whilst children with RD presented reduced scores both on the quality and the speed scales, children with DCD only appeared to be impaired in the quality scale. This result may reflect distinct underlying neuromotor mechanisms, and/or different strategies adopted during handwriting by RD and DCD children when completing the BHK test.

Children presenting with both RD and DCD obtained the worst scores in both BHK scales. They were the most impaired because they wrote slowly (like RD children) and with a decreased quality and legibility (like DCD children, and with lower quality than RD children). However, they did not differ from the singular RD for the velocity, and from the singular DCD for the quality. According to our hypothesis, our results revealed that the comorbidity of these disorders adds to, but does not worsen, handwriting deficits. This additive effect is comparable to the one observed by Downing (2018), with children adding the difficulties associated with each of the disorders when they have a DCD and a RD. However, as children of the comorbid condition probably miss resources to overcome their difficulties, we expected these children to more often reach the clinical threshold for dysgraphia. From a clinical point of view, the number of children reaching this threshold did not differ significantly between the 3 groups. However, dysgraphia seems to affect a larger proportion of children with comorbid DCD and RD (68,7%) than children with DCD (53,8%) or RD (42,1%) alone in our groups, suggesting that the consequences of these disorders may be additive. Children with comorbid RD and DCD deal with both motor and linguistic difficulties, and thus would surely find fewer available resources than children with a singular disorder. From this point of view, the consequence of the comorbid condition on handwriting would rely on a synergistic

interaction between DCD and RD, making the children more likely to fail the BHK test (Schoemaker et al., 2013).

Toward Subtypes of Dysgraphia: Comparison Between Children with DCD and/or RD

The digital analysis of the BHK produced by children with DCD or RD was considered as a mean to better understand the commonalities and differences between handwriting difficulties in these two disorders. Furthermore, the comparison between children with singular or comorbid conditions should help to better understand the developmental trajectory of the disorders and their underlying mechanisms. When focusing on children with dysgraphia, we observed that the BHK scores did not differ between the neurodevelopmental disorders. The digital analysis of handwriting helped to go beyond the paper and pen approach of dysgraphia and disentangle the contribution of the children's neurodevelopmental disorders. Thus, we observed that dysgraphic children with singular RD differed from dysgraphic children with singular DCD on four kinematic variables: tracing duration, total lift duration, total stop duration, and SNvpd. In other words, dysgraphic children with RD wrote over a longer time, with a slower velocity, and had a jerkier movement with more or longer stops than dysgraphic children with DCD. In sum, children with RD were not only perturbed by the reading of the text to copy but were also slower and less fluent. Their handwriting processes resemble students who are just beginning to learn to write, i.e. based on a visual control that prevents them from writing faster, as suggested by Barnett et al. (2019). Concerning dysgraphic children with a DCD, the paper-pen test demonstrated that they wrote as poorly and as slowly as dysgraphic children with RD, but the processes underlying their handwriting were closer to ballistic movements than dysgraphic children with RD (higher speed, smaller tracing duration, smoother movement). According to the internal modelling deficit hypothesis in DCD (Adams et al., 2014), our results suggest that children with DCD tend to be overconfident in the predictions they make from the efference copy, which

decreases sensory processing of reafferent information and prevents them from updating the existing forward model from the online control of the pen trajectory. Consequently, their movements are faster but inaccurate.

Overall, our results confirm that handwriting difficulties of children with RD and DCD rely, at least partly, on different underlying mechanisms and are interesting regarding the existing literature. They highlight that DCD primarily affects handwriting quality (Huau et al., 2015; Prunty & Barnett, 2020; Schoemaker et al., 2013) while RD affects both speed and, to a lesser extent, handwriting quality (Gosse & Van Reybroeck, 2020; Sumner et al., 2013). However, we did not report more frequent and longer pauses from children with DCD (Prunty et al., 2013, 2014). This discrepancy may be explained by the potential presence of comorbid RD within Prunty et al.'s group of children with DCD, as suggested by the fact that almost a third of the children within this group presented spelling difficulties (Prunty et al., 2013). In addition, our results bring clues to the understanding of handwriting difficulties in the context of DCD or RD alone and reinforce the idea that the handwriting difficulties observed in these two disorders are related to distinct handwriting impairments.

Finally, the digital analysis showed that at the process level, dysgraphic children with RD+DCD only differed from dysgraphic children with DCD. They showed longer lift duration and a jerkier movement than dysgraphic children with DCD. As the dysgraphic children with RD, and unlike their DCD peers, their handwriting looked like a beginner's. In sum, the results of the group comparisons suggest that dysgraphic children with both DCD and RD resemble more RD children than DCD children. The digital analysis also confirmed that comorbidity did not induce a synergistic effect, i.e. increased the severity of the dysgraphia. The dysgraphic children with RD+DCD never presented poorer performances than dysgraphic children with RD who presented the most impaired movement automation. Finally, it is noteworthy that dysgraphic children with comorbid RD and DCD showed a much

higher within-group variability than their peers. This high variability probably contributes to the small number of significant differences between the groups with dysgraphia in the analysis of the digital variables. This variability deserves more investigation as it might hide two or more groups of children. The first hypothesis is that some children in this group would be more greatly impaired than others, confirming that the spread of deficits causing the comorbidity could also increase the severity of symptoms (Schoemaker et al., 2013). Another hypothesis is that some children of the RD+DCD group present more 'DCD-like' perturbations and others present more 'RD-like' perturbations. This point of view is compatible with Jover and Huau's (2021) observation according to which both reading and motor skills explained the children's graphomotor production and with the multiple deficit model (McGrath et al., 2020; Pennington, 2006; Pennington et al., 2019). From this point of view, the coexisting risk factors would be various and of different strengths to induce comorbid RD and DCD. This would lead to different profiles of comorbid children: the 'DCD-like' and the 'RD-like'.

Limitations

The main limitation of our study concerns the sample size, which prevents us from going deeper into the characterization of different profiles within each neurodevelopmental disorder, whereas different subtypes of DCD (e.g., Vaivre-Douret et al., 2011), different subtypes of RD (e.g., Castles & Coltheart, 1993), and probably different subtypes of comorbid children, exist. A large-scale study would reveal the extent to which different subtypes of each neurodevelopmental disorder can lead to different types of dysgraphia. For instance, it has been suggested that different types of RD may lead to different types of dysgraphia (Zoccolotti et al., 2010). To what extent one of these two types of RD leads to writing performance closer to that of DCD children constitutes a first perspective.

Another limitation of this study is that the BHK test is a copy task that involves reading time, which needs to be taken into account to avoid the child's slowness in writing being the result of this increased reading time. Performing this task on a graphics tablet enables us to identify pen lifts that are considered key moments for looking at the model to be copied. Nevertheless, the addition of eye-tracking would allow reading times to be better taken into account. Furthermore, our finding based on a copy task must be confirmed in a free production writing task to be replicated and generalized.

Finally, producing the writing task when placing paper over a graphic tablet may have a double constraint that needs to be considered: On the one hand, the presence of the tablet may affect the performance, as the child is placed in an unusual condition. On the other hand, to our knowledge, the potential for loss of digital measurement accuracy when placing paper over a graphic tablet has never been investigated, and could be significant in some cases.

Conclusions

This study provides indications to help the understanding of handwriting deficits in the context of either DCD or RD and supports the hypothesis that handwriting difficulties are related to distinct neuromotor impairments. Note that this study does not allow us to determine whether the deterioration in handwriting movements in children with RD results from a motor deficit associated with possible dyslexia (in line with the cerebellar deficit hypothesis, Nicolson et al., 2001; see also Alamargot et al., 2020), or from a linguistic deficit impacting the motor control of handwriting movements, given that motor and orthographic processes interact (e.g., Danna et al., 2022; Kandel & Perret, 2015). These observations highlight the importance of performing a complete diagnosis of children, including an assessment of both motor and reading skills. They also give clues to potential intervention strategies to remediate handwriting deficits in children with RD, DCD, or RD+DCD. For

instance, our study confirms that the slowness of writing in children with DCD is not linked to their average pen speed, but probably to macrography, i.e., the tendency to write bigger, or supernumerary strokes. Consequently, a rehabilitation strategy based on the addition of spatial constraints would be more relevant than one based on the addition of temporal constraints (e.g., instruction to write faster) for children with DCD, while the reverse might be more relevant with children with RD. This study is important not only from a clinical perspective for the subsequent rehabilitation of these children by occupational therapists but also for the inclusion of children in research cohorts. The identification of subtypes of dysgraphia requires a fine-grained analysis of the handwriting process and product. In clinical practice, the test scores are not sufficient to disentangle the various difficulties. It is thus important to develop automated tools to assess handwriting at both levels for the identification of dysgraphia (Asselborn et al., 2018; Deschamps et al., 2021; Dimauro et al., 2020; Sihwi et al., 2019).

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