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► **To cite this version:**

Marianne Jover, Andréa Huau. A Dimensional Approach to Comorbidity Between Developmental Coordination Disorder and Dyslexia: A Study of Graphomotor Abilities. *A.N.A.E. Approche neuropsychologique des apprentissages chez l'enfant*, 2021, 175 (33), pp.673-688. hal-03874500

HAL Id: hal-03874500

<https://hal.science/hal-03874500>

Submitted on 28 Nov 2022

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Jover, M., Huau, A (2021). Approche dimensionnelle de la comorbidité entre trouble développemental de la coordination et trouble spécifique de l'apprentissage de la lecture : étude sur la graphomotricité/ A Dimensional Approach to Comorbidity Between Developmental Coordination Disorder and Dyslexia: A Study of Graphomotor Abilities. *Approche Neuropsychologique des Apprentissages chez l'Enfant*, 175(33), 673-688.

A Dimensional Approach to Comorbidity Between Developmental Coordination Disorder and Dyslexia: A Study of Graphomotor Abilities

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Abstract

The present study examined the comorbidity between developmental coordination disorder and dyslexia, adopting a dimensional approach. Participants were 120 children aged 8-12 years, with or without developmental disorders. After assessment with a reading test (Alouette) and motor test (MABC), children performed handwriting and graphomotor tasks on a graphic tablet. GAM analyses were used to test the contribution of the children's reading and motor test scores to their handwriting and graphomotor performances. Results revealed a correlation between reading and motor scores, despite considerable heterogeneity within the groups. The variables measured during the handwriting and graphomotor tasks often depended on the children's scores on the motor and reading tests. Results support the hypothesis of shared risk factors for DCD and dyslexia, and confirm the relevance of adopting a dimensional approach to neurodevelopmental disorders.

Keywords: developmental coordination disorder, dyslexia, comorbidity, neurodevelopmental disorders, graphomotor abilities, dimensional model

A Dimensional Approach to Comorbidity Between Developmental Coordination Disorder and Dyslexia: A Study of Graphomotor Abilities

Comorbidity refers to the co-occurrence of two diseases within the same patient (Feinstein, 1970). In the case of developmental disorders, it is commonly described as *the rule rather than the exception*. A number of clinicians and researchers have tackled this challenging subject in the past decade (Kaplan, Dewey, Crawford, & Wilson, 2001; Pennington, 2008; Smits-Engelsman, Jover, Green, Ferguson, & Wilson, 2017).

Frequent comorbidity between two given disorders raises questions about the links between their etiology and pathogenesis, as well as about the quality of their diagnostic classifications (Angold et al., 1999). According to this view, “comorbidity may not imply the presence of multiple disorders or dysfunctions but could reflect our inability to apply a single diagnosis to account for all symptoms” (Dewey, 2018, p. 5). Disorders should therefore be regarded as patterns of underlying impairment on a continuum or set of dimensions, rather than distinct entities or categories. We can thus make a distinction between categorical and dimensional approaches to developmental disorders (Sonuga-Barke, 1998). The former, classic approach is commonly employed by clinicians using DSM (American Psychiatric Association, 2013) or ICD (World Health Organization, 2019-2021) criteria. The latter can best be described as revolutionary (Sonuga-Barke, 2020). For instance, Thapar et al. (2017) recommend modeling neurodevelopmental disorders as *traits*, to account for the considerable heterogeneity in clinical profiles, and the overlap and comorbidity between diagnoses. For example, inattention and impulsivity, assessed according to DSM-5 criteria for attention deficit hyperactivity disorder (ADHD), could constitute a *trait*. More recently, the Research Domain Criteria project set up a cross-diagnostic and dimensional program to document and understand typical and atypical neurodevelopment. By transcending existing diagnostic boundaries, its researchers aim to identify the fundamental components of complex clinical

profiles, predict development trajectories, and guide individualized intervention approaches (Cuthbert, 2021; Licari et al., 2019). Current classifications are still mainly based on the categorical approach, but also incorporate elements of the dimensional approach. For example, in the DSM-5 (APA, 2013), *specific learning disorders* are no longer divided into three diagnostic categories (reading disorder, mathematics disorder, disorder of written expression; APA, 1994) as they were in the DSM-IV, but are viewed as being on a single dimension with *specifiers* of learning impairment in three domains: reading, writing, and mathematics (Peterson et al, 2021).

The work carried out by Pennington and his team around the *multiple deficit model*, recently renamed the *multiple factor model*, is part of this dimensional approach (McGrath, Peterson, & Pennington, 2020; Pennington, 2006; Pennington, McGrath, & Peterson, 2019). This model rejects the classic hypothesis of a single deficit explaining a given neurodevelopmental disorder. Instead, it postulates that the etiology of neurodevelopmental disorders lies at the intersection of multiple genetic and/or environmental risk and protective factors (susceptibility locus on a chromosome, premature birth, family stimulation, etc.). These shape the development of the central nervous system, the mediator of cognitive functions, and can trigger a cascade of behavioral disorders. Given the interactions between risk and protective factors, the plasticity of the central nervous system, and compensation between cognitive modules, a single risk factor cannot give rise to a developmental disorder. For the authors, there are necessarily several, possibly interrelated, predictors that contribute in a probabilistic (and not deterministic) way to the expression of a given neurodevelopmental disorder. Several risk factors must therefore coexist for a neurodevelopmental disorder to appear. This model explains both the comorbidity and the variety of clinical profiles and subtypes in neurodevelopmental disorders. Finally, this model is compatible with the hypothesis of a continuum of severity, whereby a disorder appearing in a severe form is

frequently associated with another comorbid disorder (Jongmans, Smits-Engelsman, & Schoemaker 2003; Kaplan, Crawford, Cantell, Kooistra, & Dewey, 2006; Schoemaker, Lingam, Jongmans, van Heuvelen, & Emond, 2013).

The comorbidity between developmental coordination disorder (DCD) and dyslexia (DYS) is an extensively documented example of comorbidity between neurodevelopmental disorders. It concerns 10-70% of children with DYS, and 29-70% of children with DCD (for a review, see Jover et al., 2013). Beyond the epidemiological research, many studies have specifically examined motor impairments in children with DYS, and reading impairments in children with DCD. Their results show that children with DYS frequently (but not always) exhibit motor difficulties in postural, motor learning and coordination tasks (e.g., Fawcett, Nicolson, & Dean, 1996; Haslum & Miles, 2007; Moore, Brown, Markee, Theberge, & Zvi, 1995). These motor difficulties are often specific, and rarely affect all motor areas. By the same token, children with DCD regularly have a lower reading level than control children, even if it does not reach the threshold required for a diagnosis of DYS (e.g., Alloway & Temple, 2007; Dewey, Kaplan, Crawford, & Wilson, 2002; Kadesjö & Gillberg, 1999). Despite discrepancies between research results (Chaix et al., 2007; Rochelle & Talcott, 2006) and the heterogeneity within neurodevelopmental disorders (e.g., for DYS: Norton et al., 2014; Zoubinetzky, Bielle, & Valdois, 2014; and for DCD: Gomez, & Sirigu, 2015; Tallet, & Wilson, 2020), some authors have suggested that DCD and DYS share a partly common etiology (Kaplan, Wilson, Dewey, & Crawford, 1998; Nicolson & Fawcett, 2007). The methodologies required to examine this hypothesis are complex. Several recent studies have compared the performances of children with DYS, or DCD, or both (DYSDCD) in the same task. The performances of the children in each group were analyzed to identify commonalities and specificities linked to each diagnosis (Geuze & Kalverboer, 1994), and the characteristics of the children with DYSDCD were analyzed to check whether they resembled children with

DCD or children with DYS, presented a more severe condition, or even exhibited a different profile. The results of these studies showed that children with DYSDCD sometimes have a profile similar to that of children with DYS (Biotteau, Chaix, & Albaret, 2015), or that of children with DCD (Bellocchi et al., 2017), and sometimes exhibit the difficulties of both these groups (Bellocchi, Ducrot, Tallet, Jucla, & Jover, 2021; Biotteau et al., 2017, Downing & Caravolas, 2020; Ho, Chan, Leung, Lee, & Tsang, 2005; Maziero et al., 2020). Children with comorbid DYS and DCD sometimes have greater difficulties than children with just one disorder (Cignetti et al., 2018; Jongmans et al., 2003), but this is not always the case (Bellocchi et al., 2021; Maziero et al., 2020). However, the classic categorical approach on which this research is based has the disadvantage of relying on strict inclusion criteria and of focusing on children who are often relatively unrepresentative of the clinical population, possibly explaining the contradictory results.

The aim of the present research was to document the comorbidity between DYS and DCD in an innovative way, using a dimensional approach. Visser et al. (2020) recently showed that, depending on whether a categorical or a dimensional approach is adopted, the study of the comorbidity between ADHD and DCD can yield different results. We therefore focused on graphomotor and handwriting difficulties, which are frequently reported in both children with DYS (e.g., Afonso, Suárez-Coalla, & Cuetos, 2020; Alamargot, Morin, & Simard-Dupuis, 2020; Berninger, Nielsen, Abbott, Wijsman, & Raskind, 2008; Germano, Giaconi, & Capellini, 2016; Gosse & van Reybroeck, 2020; Iversen Berg, Ellertsen, & Tønnessen, 2005, Lam Au, Leung, & Li-Tsang, 2011; Pagliarini et al., 2015,) and children with DCD (e.g., Huau, Velay, & Jover, 2015; Jolly & Gentaz, 2014; O'Hare & Khalid, 2002; Prunty & Barnett, 2020; Prunty, Barnett, Wilmut, & Plumb, 2013, 2014; Rosenblum & Livneh-Zirinski, 2008; Rosenblum, Margieh, & Engel-Yeger, 2013; Smits-Engelsman, Wilson, Westenberg, & Duysens, 2003). A literature review revealed deficits in the quality of

the writing: both the process, (i.e., way of producing the gesture) and the end-product in children with DCD or DYS (Rosenblum, Weiss, & Parush, 2003). At the product level, children with DCD exhibited letter malformation and macrography, which was not the case for children with DYS. At the process level, studies based on graphic tablet recordings showed that children with DYS or DCD had a lower writing speed than controls, mainly owing to more writing pauses (Prunty, Barnett, Wilmut, & Plumb, 2013; Sumner, Connelly, & Barnett, 2013). Children with DCD had significantly lower handwriting fluency than that of typically developing children, with more spikes in speed. This variable was not measured in studies among children with DYS. Finally, significant intra-individual variability was found in both disorders. In sum, graphomotor dysfunctions were detected in both children with DCD and children with DYS. The remaining question concerns the origin of these dysfunctions, and their similarity or dissimilarity in these two neurodevelopmental disorders.

According to Downing (2018), the graphomotor difficulties of children with DYS are mainly due to fluency problems, whereas those of children with DCD arise from problems of letter formation (legibility). The reduced fluency in DYS, reflected in longer pauses during the handwriting process, can be explained by slow decoding of the words increasing working memory load and by the visual back and forth between the model and the sheet when copying. Handwriting difficulties are therefore the result of spelling and reading difficulties here (Arfé, Corato, Pizzocaro, & Merella, 2020; Martínez-García, Afonso, Cuetos, & Suárez-Coalla, 2021; Suárez-Coalla, Afonso, Martínez-García, & Cuetos, 2020; Sumner, Connelly, & Barnett, 2014). By contrast, children with DCD tend to have difficulty setting up the handwriting motor program and performing the graphic movement (Barnett & Prunty, 2020; Huau et al., 2015; Jolly, Danna, & Jover, in preparation).

The aim of the present research was to use a dimensional approach to improve our understanding of the comorbidity between DCD and DYS, as well as the writing difficulties

observed in children with these disorders. Accordingly, after recruiting a group of children with DCD, DYS, or both (DYSDCD), together with a group of age-matched controls (CTL), we administered standardized tests to measure their levels of reading and motor skills. Based on the multiple factor model, we expected children's motor and reading skills to lie on a continuum of impairment, with considerable heterogeneity in performances on both tests (Pennington, 2006; Pennington et al., 2019). In addition, we administered a graphomotor precision task and a word writing task with or without speed constraint, as well as a writing test (BHK; Hamstra-Bletz, de Bie, & Berry, 1987). The aim was to measure the proportion of variance in performances on the graphomotor and writing tasks (process and product) explained by the children's reading and motor skills scores. To this end, we calculated generalized additive models (GAMs), which investigate nonlinear relationships and interactions between variables without a priori assumptions on their form. We expected the children's motor level to contribute more than their reading level to the variability in performances, especially on the graphomotor (vs. handwriting) tasks. We expected children with a low reading level to make longer pauses in writing tasks. Finally, performances on long or constrained tasks would depend more heavily on the children's motor skills and reading performances. The variables related to the writing product would be impacted more by motor performance, and those related to the writing process by reading performance, at least when these performances were impaired. The dimensional approach would therefore make it possible to identify the specific and possibly nonlinear effects of reading and motor difficulties associated with DYS and/or DCD on graphomotor and handwriting performances.

Methods

Participants

Participants were 130 children (Table 1), divided into two groups. The first group ($n = 65$ children, age range: 8-12 years) had exhibited difficulty acquiring reading and/or

coordination, leading professionals to establish a diagnosis of DYS and/or DCD with their usual tools (DYS, DCD and DYSDCD groups). All the children were receiving treatment for one and/or the other of these disorders. They were contacted through a professional network (psychomotor therapists, speech therapists) or through a local learning disability reference center (Timone Marseille Hospital, Aix-en-Provence Hospital). We did not control for the presence of other comorbidities, except for ADHD. Children with ADHD, together with those who exhibited excessively impaired graphomotor skills, (spontaneous capital letter writing) were excluded from the sample.

The second group ($n = 65$) comprised children matched for age who had never attended a consultation for developmental difficulties (CTL group). These children were recruited from a primary school and a junior high school in the Marseille region. Children who had repeated a grade were excluded from the sample. A questionnaire attached to the information and consent form that was sent to parents allowed us to identify the children who needed to undergo a speech and/or psychomotor assessment. These children were excluded from the sample.

This study was carried out in accordance with the Declaration of Helsinki, and parents and children all signed a consent form after being informed of the study's objectives and content.

Procedure and Material

All the children in the final sample were assessed with the Alouette reading test and the Movement Assessment Battery for Children (MABC; Henderson & Sugden, 1992), and performed graphomotor and handwriting tasks. The study took place either in the laboratory or at the children's school.

The French adaptation (Soppelsa & Albaret, 2004) of the MABC was used to assess the motor skills of children aged 4-12 years in three domains: manual dexterity, ball skills, and static and dynamic balance. This battery contains items specific to four different age groups (4-6 years, 7-8 years, 9-10 years, 11-12 years). For each item, the instruction is accompanied by a demonstration and then a training trial, followed by formal trials. The test yields a score for each domain and a total impairment score. In the present study, the total score, expressed as a percentile, was used to characterize each child's level of motor performance.

The Alouette reading test (Lefavrais, 1965; revised version, 2005) is widely used in France to assess the accuracy and speed of reading aloud in children from elementary school to middle school. The text comprises 265 words, and the reading time is limited to 3 minutes. The sentences are not integrated into an overall structure such as a narrative, and drawings around the text depict words from the text or words that are phonologically related. Children are told to read the text and are informed that no comprehension questions will be asked. We used the corrected speed-accuracy reading score, which takes into account both reading accuracy and speed, and the z score was converted into a percentile to characterize each child's reading level.

The BHK is a handwriting test for children (French adaptation: Charles et al., 2003; Hamstra-Bletz, de Bie, & Berry, 1987) or adolescents (Soppelsa & Albaret, 2013) that consists in copying a text for 5 minutes. The first five sentences comprise monosyllabic words encountered in first grade, but the text then becomes more complex, and the size of the characters decreases. It is copied on a white unlined A4 sheet. Writing quality is scored on a set of criteria (max. 13 for the Child version; 9 for the Adolescent version). A speed score is also calculated from the number of letters copied in 5 minutes. The quality and speed scores were z -transformed according to age and sex, and used as the dependent variables.

The graphomotor control task consisted in tracing the displacement of a mouse toward a piece of cheese (Fig. 1). The first step was to connect five targets measuring 2.5 x 3 mm that were spaced 50 mm apart, and the second was to draw a line between two lines spaced 2 mm apart. The model was printed on a sheet of paper positioned on the graphic tablet. This task was performed either at the speed chosen by the children (normal speed condition) or as quickly as possible (fast speed condition). The numbers of missed targets and line crossings were summed to use as a dependent variable (Precision).

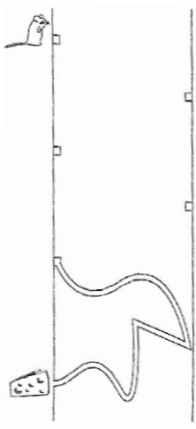


Figure 1. Model of the graphomotor control task.

The word writing task consisted in writing the word *Lapin* [Rabbit]. The word (28 x 20 mm) was printed in cursive letters on a sheet of paper placed next to the tablet. The children had to write in 57 x 19-mm preprinted boxes, on a sheet of paper positioned on the tablet. They performed eight trials at "usual speed", then eight at fast speed "as quickly as possible". The number of trials with erasures or corrections was counted and used as a dependent variable (*corrected trials*).

The graphomotor control task and the *Lapin* word writing task were performed on a Wacom Intuos 4L graphic tablet. Its A4 format recording surface (29.7 x 21 cm) allows pen movements to be recorded with good spatial (0.25 mm) and temporal (200 points / s) precision. Children wrote with an Inking Pen, a ballpoint pen of classic size and heft. The tablet was placed on the school desk in the landscape orientation. Data processing was carried

out with the Ecriture Suite 2 software (Gilhodes & Velay, 2012). The Segment Run tool was used offline to clean up the recorded data. The Stats Trace tool allowed us to extract various indices from the recordings. The variables recorded by the tablet and used in this study were 1) *trace length* (mm; i.e., length of the child's production), where the trace is unwound like a thread to provide an index of production, 2) *mean velocity* (mm / s), a kinematic indicator of the writing movement, 3) *pause duration* (s), namely the total time spent still, without moving or lifting the pen, accounting for lack of fluency in the movement.

The *precision*, *corrected trials* and *trace length* variables provided indices of the result/ product of the graphomotor movement. The *mean velocity* and *stop duration* variables accounted for the motor processes during task performance.

Statistical Analysis

To describe the sample, we compared the scores of the children of the CTL, DCD, DYS and DYSDCD groups) with a Kruskal-Wallis ANOVA, and carried out post hoc analyses using the Dwass-Steel-Critchlow-Fligner test.

The following analyses were performed on the data of the whole sample. The motor and reading assessment scores were subjected to a correlation analysis (Spearman's rho). Generalized additive models (GAMs) were used to identify the contributions of motor skills and reading performance (as assessed by the MABC and the Alouette), and their possible interaction, to children's graphic and writing tasks. GAMs make it possible to model the link between a variable to be explained and several continuous predictors. The objective is to maximize the prediction of a dependent variable from several predictors and achieve the best fit to real-world data by considering the possible nonlinear relationships between explanatory variables. An F test is used to gauge the significance of the contribution of the explanatory variables ($p < .05$), and R^2 indicates the variance explained by the model. Graphical

representations are provided to interpret the effects of the predictors. They make it possible to identify whether the interaction effects are absent, simple, linear, or nonlinear, based on the color gradient and the orientation of the isolines representing the linear term. The standard error is shown in dotted lines on the graphs, to indicate prediction quality. The GAMs were estimated using the *mgcv* package in R (Woods, 2021).

Results

Alouette and MABC Scores

Table 1 summarizes the scores of the children in the four groups and the results of the intergroup comparisons. The group effect was significant for both the MABC ($\chi^2 = 61.5, p < .001, \varepsilon^2 = .477$) and Alouette ($\chi^2 = 67.9, p < .001, \varepsilon^2 = .526$) scores. Paired comparisons carried out with the Dwass-Steel-Critchlow-Fligner test showed that the differences between the groups were always significant, except between the DYSDCD and DCD groups for the MABC score, and between the DYSDCD and DYS groups for the Alouette score.

Table 1

Group Comparisons on MABC and Alouette Scores

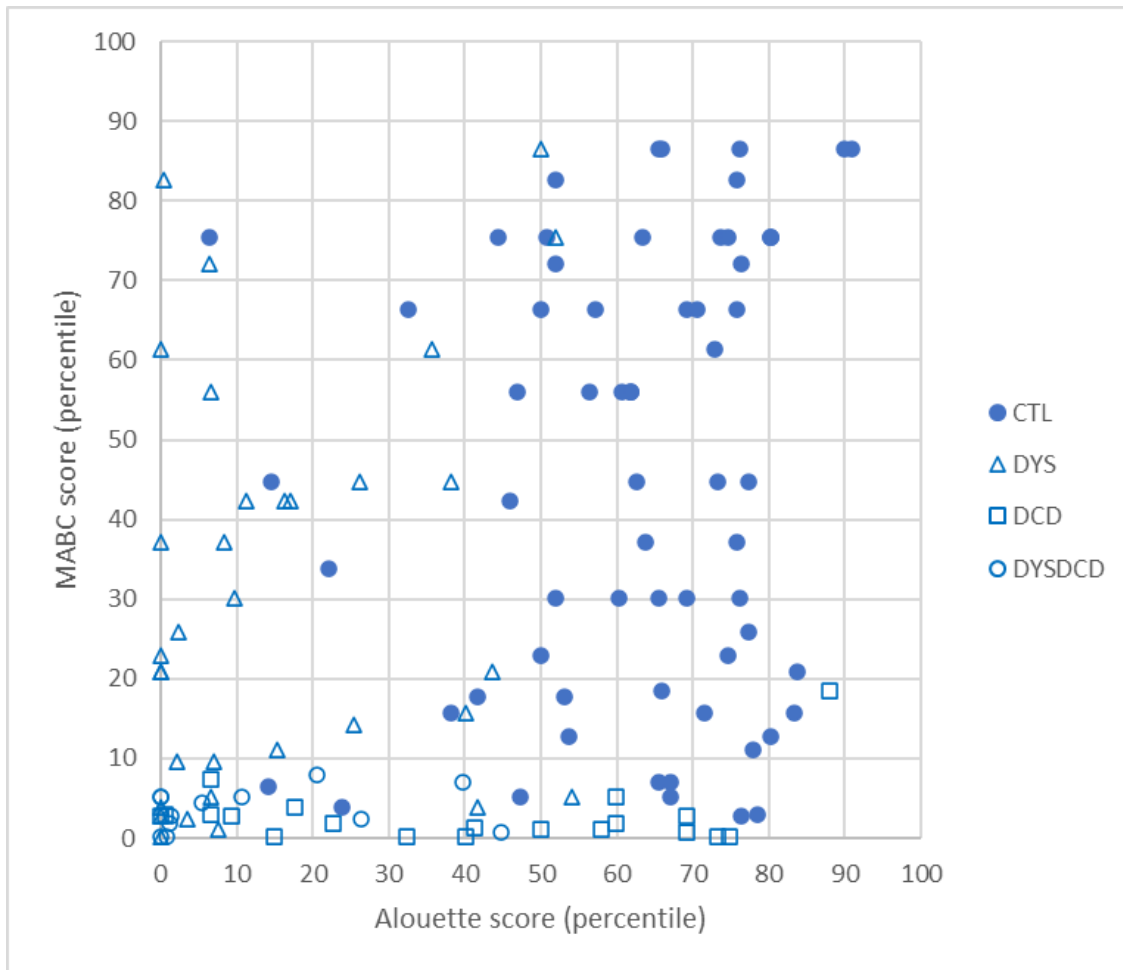
Group	<i>n</i> (girls)	Mean age (<i>SD</i>)	MABC mean percentile (<i>SD</i>)	Alouette mean percentile (<i>SD</i>)
CTL	65 (28)	115 (13)	45.6 (27.7)	61.9 (18.5)
DYS	33 (10)	119 (14)	30.7 (25.7)	16 (18)
DYSDCD	12 (3)	118 (14)	3.57 (2.63)	12.6 (16.4)
DCD	20 (4)	115 (13)	2.86 (4.09)	39.7 (28.4)

<i>ns</i>	CTL>DYS*	CTL>DYS***
	CTL>DYSDCD***	CTL>DYSDCD***
	CTL>DCD***	CTL>DCD**
	DYS>DCD***	DCD>DYS*
	DYS>DYSDCD**	DCD>DYSDCD*
	DYSDCD; DCD	DYSDCD; DYS

Note. DYS: dyslexia; DCD: developmental coordination disorder; DYSDCD: DCD + DYS; CTL: controls; *SD*: standard deviation; *ns*: nonsignificant. * $p < 0.05$. ** $p < 0.01$. *** $p < 0.01$.

Figure 2 shows the children's distribution according to their scores on the MABC and Alouette tests. Correlations between scores were significant ($\rho = .34, p < .001$).

Figure 2. Participants' MABC and Alouette scores.



Note. DYS dyslexia; DCD: developmental coordination disorder; DYSDCD: DCD + DYS; CTL: control.

Alouette and MABC scores below the 15th percentile are usually considered to indicate a risk of the relevant disorder, and scores below the 5th percentile a proven disorder. Here, five children in the CTL group and eight children in the DYS group were below the 5th percentile on the MABC test, as were two children in the DCD group on the Alouette test. A total of 15 children in the CTL group and 13 children in the DYS group were below the 15th percentile on the MABC, as were three children in the CTL group and six children in the DCD group on the Alouette.

GAM Analyses: Effects of Reading and Motor Skills Levels on Graphomotor Dependent Variables

In the graphs illustrating the significant GAM models (Figs. 3, 4, 5 and 6) reading level (Alouette scores as percentiles) is on the x-axis, and motor skills level (MABC scores as percentiles) is on the y-axis. The black dots represent the children in the sample. In the heat maps, the color gradient corresponds to the values of the dependent variable predicted by the model. The lighter the color, the higher the predicted value, and the darker the color, the lower the value. For example, in Figure 3, a high number of missed targets is shown in pale yellow, and a low number is shown in red. The green lines are isolines indicating the predictor gradients, where 0 represents the mean. They can be either rectilinear, indicating a linear effect of reading and motor skills scores, or curved, indicating a nonlinear effect of potentiation of one explanatory variable by the other. When the lines are oblique, the effects of reading level and motor skills were additive. When the lines are vertical, reading had a predominant effect, and when they are horizontal, motor skills had a predominant effect. A second type of model presentation illustrates the variability of the GAM estimates. The black lines represent the main estimates, and the red and green dashed lines -1 and +1 standard error respectively.

Contributions of motor skills and reading levels to graphomotor control task.

There was an additive linear effect of reading and motor skills levels on graphomotor performances. The lower children's reading and motor skills levels, the greater the numbers of they missed targets and line crossings, in both normal and fast speed conditions (Fig. 3). This effect was also observed for mean velocity (Fig. 4), as the better the children's reading and motor skills, the faster they completed the task. Finally, in both models, the fast speed condition clearly increased the variance explained by the model.

	Normal speed	Fast speed
<i>t</i> value (Intercept)	10.17***	13.67***
<i>F</i>	9.278***	8.177***
<i>R</i> ²	16.5%	26.2%

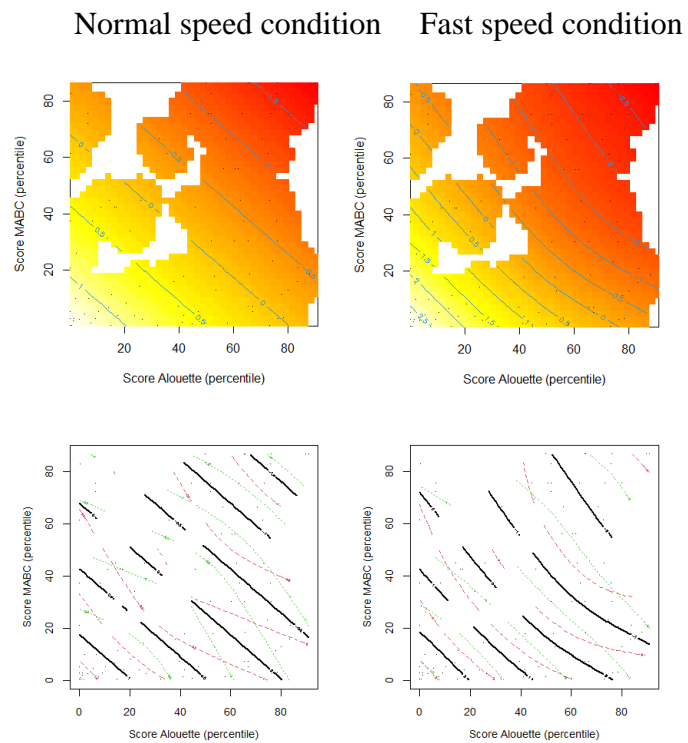


Figure 3. Precision in the graphomotor control task: statistical values and graphical representations of the GAM model for the normal speed and fast speed conditions: heat map and variability. *** $p < 0.001$.

	Normal speed	Fast speed
<i>t</i> value (Intercept)	39.9***	33.05***
<i>F</i>	4.37*	6.025**
<i>R</i> ²	8.51%	11.5%

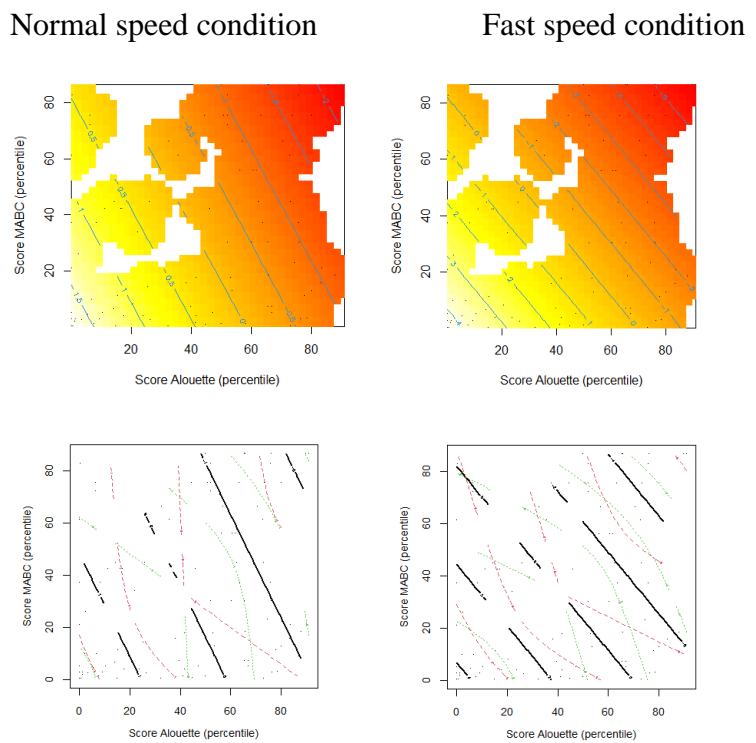


Figure 4. Mean velocity in the graphomotor control task: statistical values and graphical representations of the GAM model for the normal speed and fast speed conditions: heat map and variability. * $p < 0.05$. ** $p < 0.01$. *** $p < 0.001$.

Contribution of motor skills and reading levels to word writing task.

The GAM model applied to the parameters recorded when writing the word *Lapin* at normal speed did not reveal a significant effect of either motor skills or reading levels. By contrast, in the fast speed condition, several effects appeared (Fig. 5). The graphs show a simple effect of reading level on the number of corrected trials, such that the children with more severe reading difficulties had more corrected trials. In addition, there was an additive effect of reading and motor skills on trace length: the more severe the children's reading and motor difficulties, the longer their trace. Pause duration was sensitive to both reading and motor skills levels: children with a low reading level made longer pauses when they had a good level of motor skills. Good readers with severe motor difficulties spent less time pausing. Writing velocity, by contrast, did not appear to be sensitive to either motor skills or reading levels.

	Corrected trials	Velocity	Trace length	Pause duration
<i>t</i> value (intercept)	5.486***	33.55***	35.1***	8.67***
<i>F</i>	4.411*	3.026	4.782*	3.15*
<i>R</i> ²	7.18%	5.08%	7.81%	5.37%

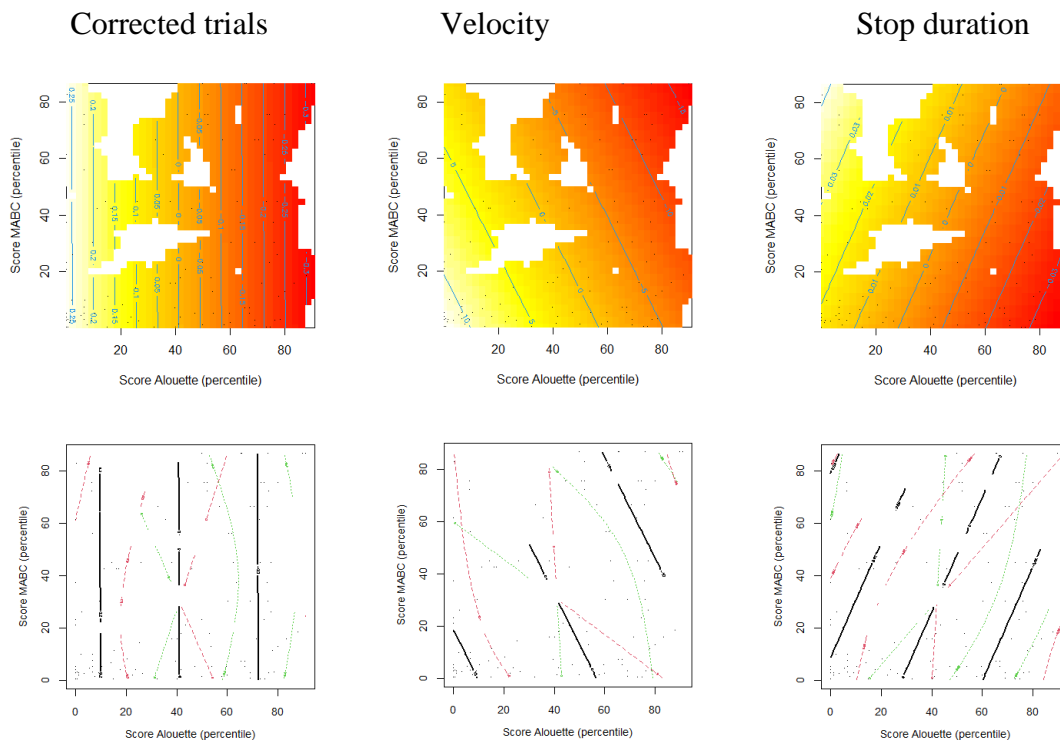


Figure 5. Parameters recorded for the word *Lapin* in the fast speed condition: statistical values and graphical representations of the GAM model: heat map and variability. * $p < 0.05$. *** $p < 0.001$.

Contributions of motor skills and reading levels to BHK scores.

Regarding BHK scores, the GAM was significant both for quality and for speed (Fig. 6). There was an interaction between the influence of reading and motor skills levels on the quality score. This effect was linear when children had low motor and reading skills, such that the more difficulties they had, the lower the quality. In addition, children with low motor skills had a low score, regardless of their reading level. The effect became nonlinear when

children had average-to-high levels of motor and reading skills. In this case, children produced high-quality writing and there was a simple effect of reading.

Concerning the BHK speed score, we observed a simple effect of reading (Fig. 6), such that the lower the children’s reading level, the lower their writing speed.

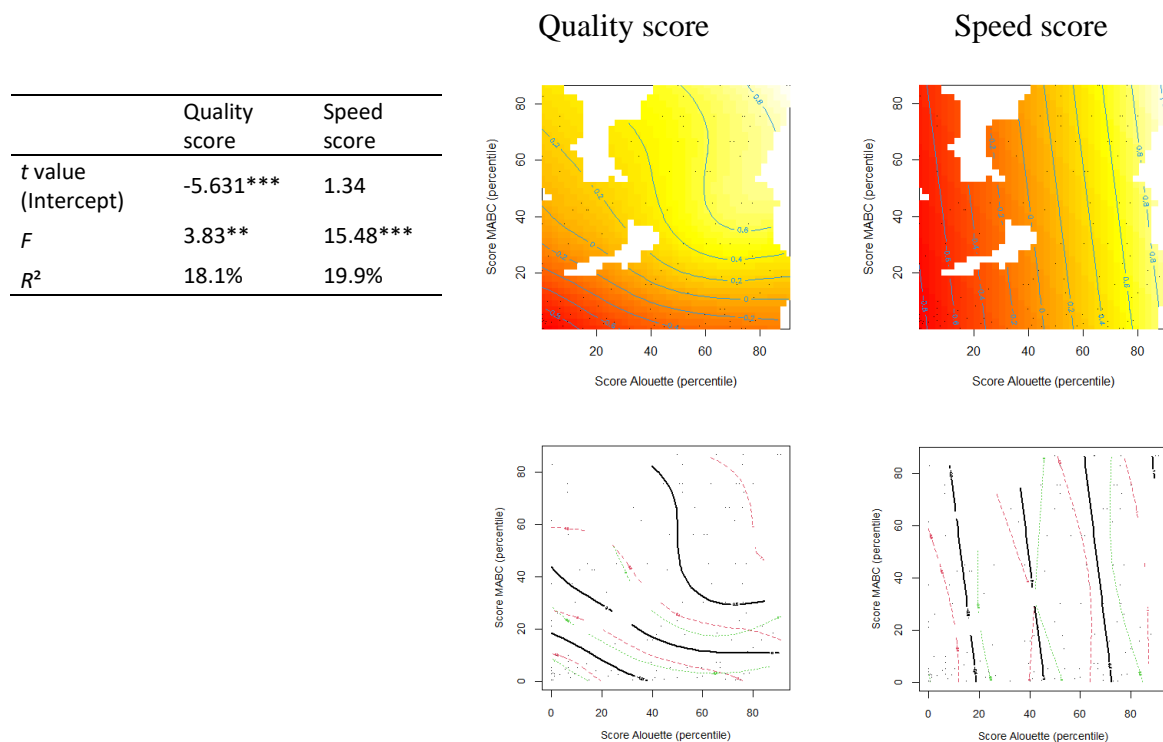


Figure 6. BHK test scores. Statistical values and graphical representations of the GAM model: heat map and variability. ** $p < 0.01$. *** $p < 0.001$.

Discussion

The aim of this research was to examine how a dimensional approach to disorders can provide a better understanding of the comorbidity between DYS and DCD and the manifestation of handwriting difficulties in these two disorders. We therefore recruited a sample of children with or without a diagnosis of DYS and/or DCD. All children were assessed on standardized motor skills (MABC; Henderson & Sugden, 1992) and reading

(Alouette & Lefavrais, 1965) tests, and we analyzed the links between their scores. We expected children in the CTL, DCD, DYS and DYSDCD groups to lie on a continuum, with children with DYSDCD displaying more impaired performances than children with just one disorder. In addition, we recorded children's graphomotor output in a graphomotor precision task, a word writing task with or without speed constraint, and a handwriting test (BHK). We then ran GAMs to test the contributions of children's reading and motor performances to the variance in measurements recorded during the graphomotor and writing tasks. We expected motor skills level to contribute more than reading level, especially to graphomotor precision. We predicted that in the writing tasks, children with a low reading level would make longer pauses. Finally, we predicted that performance on lengthy tasks or tasks with a speed constraint would depend more clearly on the children's motor skills and reading levels. In sum, the variables pertaining to the writing product or writing process would not be sensitive to motor and reading scores in the same way. Our results did not validate all these hypotheses, and even invalidated some of them.

The first interesting result of our analyses concerned children's scores on the Alouette and MABC tests. The clinical profiles of the control children and children with DCD, DYS, or DYSDCD we recruited were more similar than expected. For instance, some of the children who had no reading or coordination problems, according to their parents, had considerable difficulty performing the Alouette or MABC tests. This observation has two possible non-mutually exclusive explanations. First, some children probably have difficulties that go unnoticed by their parents or teachers. Some children also find ways of compensating for their difficulties in the course of everyday life, but these difficulties are still revealed by specific standardized tests. More children presented worrying test scores for motor difficulties, doubtless reflecting the fact that schools are far more vigilant for reading difficulties than for physical ones, which are socially better tolerated (Blank et al., 2019).

Children with reading or motor skills test scores below the 5th percentile are likely to encounter substantial difficulties at home and in school if no help is offered. This observation reinforces the need for elementary and middle schools to have qualified staff who can identify children at risk of neurodevelopmental disorders and refer them to specialists, who can then make appropriate recommendations in terms of assessment and follow-up (Haute Autorité de Santé, 2017, 2020; see also Camden et al., 2020).

Analysis of MABC and Alouette test scores across the whole sample showed that the children were distributed almost over the entire graph. Some had a very high level of motor skills and a very low or very high reading level, while others had a very high reading level and either very good or very poor motor skills. There is therefore considerable heterogeneity: not all children with DYS have a motor disturbance (Ramus, Pidgeon, & Frith, 2003), and not all children with DCD have impaired reading ability (Zwicker, Missiuna, Harris, & Boyd, 2012). However, in the present study, MABC and Alouette scores were significantly correlated. In addition, the group comparison showed that children with DYS performed more poorly on average than the CTL group on the MABC test. By contrast, children with DCD performed more poorly on average than the CTL children on the Alouette test. Children with isolated DCD or DYS therefore generally exhibit a low level of functioning in the domain not concerned by the neurodevelopmental disorder. These results confirm the severity continuum hypothesis whereby in its severe form, one disorder is more frequently associated with another one (Jongmans et al. 2003; Kaplan et al., 2006; Schoemaker et al., 2013). These elements reinforce the relevance of adopting a dimensional approach to neurodevelopmental disorders: the transitions from typical to atypical functioning, and from isolated disorder to comorbid situation occur on a continuum (Dewey, 2018; Licari et al., 2018; Pennington, 2006; Thapar et al. 2017).

GAMs provided an opportunity to analyze the respective effects of motor skills and reading on graphomotor and writing tasks. Results showed that reading and motor skills levels, respectively assessed with the Alouette (Lefavrais, 2005) and MABC (Henderson & Sugden, 1992) tests, influenced children's performance on the graphomotor control task in an additive way. Velocity and precision were higher in children with high levels of both reading and motor skills, and lower in children who displayed difficulties in both domains at the same time. Performances were average in children with poor motor skills and good reading skills, and in children with poor reading skills and good motor skills. Even though this graphomotor task did not involve language, the contribution of reading level was equivalent to that of motor skills. Insofar as earlier research had suggested that children with DYS have reduced motor abilities, this result was unexpected in a study that considered levels of both motor skills and reading, and in which we *controlled* for comorbidity. However, it corroborates recent observations showing that writing difficulties are correlated with children's reading difficulties (Arfé et al., 2020; Martínez-García et al., 2021; Suárez-Coalla et al., 2020; Sumner et al., 2014). In our opinion, it shows that this simple graphomotor task requires skills related to both reading and motor control, and which can be compensated for. However, further research needs to be undertaken to understand this result better.

When children had to write the word *Lapin* with a speed constraint, we observed specific reading level effects. The number of corrected trials depended exclusively on reading level, such that children with a low score on the Alouette (Lefavrais, 2005) made the most corrections. For their part, Sumner and Connelly (2020) found that even at university, students with dyslexia made more crossings out than their peers did. The MABC score did not influence this variable. Trace length and stop duration also depended on both reading and motor skills. However, for the latter variable, the color gradient indicated that children who paused the longest were poor readers with good motor skills. Children with poor motor skills

tended to spend less time pausing. This result is consistent with the findings of Downing (2018) and Jolly, Danna, and Jover (in preparation) for children with DYS, DCD and DYSDCD. It supports the hypothesis that children with DYS have specific writing difficulties, compared with children with DCD.

Finally, analysis of BHK scores showed that in this copy task, motor skills and reading levels both influenced children's performance. Writing quality was most impaired in children with poor motor skills, regardless of their reading level. However, this effect was nonlinear, and when children reached the average motor skills level, reading level became decisive. Being a proficient reader therefore determined writing quality when children had no motor difficulties. This result sheds light on the many determinants involved during writing, and the complex interactions between linguistic and motor skills in this activity (Gosse, et al., 2020; see also Danna, Longcamp, Nalborczyk, Velay, Commengé, & Jover, under review). Concerning writing speed, a simple reading effect was observed, whereby the lower the children's reading level, the lower their BHK score. Children with poor reading skills never had a high writing speed, whereas children with poor motor skills could write quickly if they could read well. This result should be set against the nature of the BHK test (Hamstra-Bletz, de Bie, & Berry, 1987), which is a text copy test. It is easy to see why children with reading difficulties would perform a copying task more slowly, as this task places particularly heavy demands on working memory (Sumner et al., 2014). Our results also showed that children with a low motor score were not necessarily slower than the others if they did not have to read. This result should be considered in the light of Barnett and Prunty (2020)'s recent review, which indicated that not all children with DCD are slower.

Ultimately, it is interesting to note that time pressure (or task length) in the graphomotor and writing tasks was linked to GAMs of greater explanatory power. Thus, motor skills and reading levels contributed more to the predicted variable when the situation

was constrained. This result is consistent with previous research among children with DCD or DYS, where the gap with controls widened according to the level of task constraint (Gosse & Van Reybroeck, 2020; Huau et al., 2015). It confirms the findings of clinicians and of developmental studies highlighting an increase in the difficulties of children with neurodevelopmental disorders when school and/or social constraints become more severe (Pennington et al., 2019).

The results of the present study provide a better understanding of the discrepancies between research indicating that children have more severe difficulties in the case of comorbidity (e.g., Jongmans et al., 2003) and research that does not (e.g., Maziero et al., 2020). Depending on the tasks and variables analyzed, the effects of reading and motor skills levels may either be independent, additive, or interact in a nonlinear way.

In conclusion, the indices measured in the graphomotor and writing tasks depended on children's performances on both the MABC motor skills and Alouette reading tests. The effect of reading level was comparable to that of motor skills in the graphomotor tasks, but became more salient in the writing tasks, when the children had to write at a fast speed. This study shows how important reading level is in writing tasks, and confirms more or less recent results showing a link between basic reading tasks and writing (Johannessen, Longcamp, Stuart, Thibault, & Baber, 2021; Longcamp, Anton, Roth, & Velay, 2003) or models of human cognition (see Grw factor in Cattell–Horn–Carroll model; Schneider & McGrew, 2018). However, this link is not addressed in diagnostic classifications such as DSM-5 or ICD-11, and writing difficulties are not included among the signs of specific reading impairment (Peterson et al., 2021).

The results of this research support the multiple factor model explanation for symptom overlaps and comorbidities in neurodevelopmental disorders (McGrath et al., 2020; Pennington, 2009; Pennington, et al., 2019). The existence of shared risk and protective

factors explains the presence of a continuum of severity (Jongman et al., 2003; Kaplan et al., 2006; Schoemaker, et al., 2013), where children in the greatest difficulty at the motor level, are generally also in the greatest difficulty at the reading level. GAM models helped to identify the contributions of motor skills and reading levels to task performances, and provided an innovative means of understanding comorbidity and its effects on individuals. Our results show that while most of the comorbidity effects were additive, reading and motor difficulties had distinct effects on writing pauses, attesting to the influence of specific factors (McGrath et al., 2020; Pennington, 2006).

Nevertheless, the present study had several limitations that restrict the generalization of its results. First, we used children's composite scores on a single test as an indicator of their level in reading or motor skills. This methodological bias constitutes a limitation, as no single test can capture such complex skills. For example, the Alouette test focuses on the decoding process, does not consider text comprehension, and includes drawings meant to mislead children (Lefavrais, 2005). Likewise, the MABC test is not the gold standard for motor assessment (Henderson & Sugden, 1992). These results must therefore be replicated before they can be generalized. It would also have been interesting to compare the dimensional analysis with a categorical analysis, in order to better understand the value of each approach (Visser et al., 2020). Furthermore, a latent profile analysis (e.g., Spurk, Hirschi, Wang, Valero, & Kauffeld, 2020) would have allowed us to look for different profiles, based on the children's test performances. Finally, we only described a small number of indices and graphomotor and writing tasks here. Handwriting analysis with digital tools is a useful way of identifying the handwriting parameters that are typical of a particular difficulty (Gargot et al., 2020; Palmis, Danna, Velay, & Longcamp, 2017). More research is needed to pinpoint the differences between the dysgraphic handwriting of children with DYS and the dysgraphic handwriting of children with DCD (Jolly, Danna, & Jover, in preparation).

In conclusion, we showed that the dimensional analysis of neurodevelopmental disorders provides a means of documenting their comorbidity and etiology in a complementary way. This article is part of a more general epistemological movement to redefine developmental disorders (Habib, 2021; Pennington et al., 2019; Sonuga-Barke, 2020). In our opinion, future studies should more carefully consider children's comorbidity and the variability in their clinical profiles, in order to better contribute to the understanding of the risk and protective factors involved.

Acknowledgments

The present research was supported by a grant from the French National Research Agency (ANR-13-APPR-0010-04). The authors would like to thank Jean-Luc Velay for his help with the recordings, and Bruno Dauvier for his assistance with the analyses. This study would not have been possible without the participation of the children, the consent of their parents, and the support of local teachers, psychomotor therapists, and speech therapists. We are most grateful to them all.

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