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Atypical procedural learning skills in children with Developmental Coordination Disorder

Dorine Van Dyck, Nicolas Deconinck, Alec Aeby, Simon Baijot, Nicolas Coquelet, Xavier De Tiège and Charline Urbain

ABSTRACT
We investigated the procedural learning deficit hypothesis in Developmental Coordination Disorder (DCD) while controlling for global performance such as slower reaction times (RTs) and variability. Procedural (sequence) learning was assessed in 31 children with DCD and 31 age-matched typically developing children through a serial reaction time task (SRTT). Sequential and random trial conditions were intermixed within five training epochs. Two repeated measures ANOVAs were conducted on a Sequence-Specific Learning Index (SSLI) and a Global Performance Index (GPI, speed/accuracy measure) with Epoch (for SSLI and GPI) and Condition (for GPI) as within-subjects factors, and Group as between-subjects factor. Controlling for RTs differences through normalized RTs, revealed a global reduction of SSLI in children with DCD compared with TD peers suggesting reduced sequence learning skills in DCD. Still, a significant Group x Condition interaction observed on GPI indicated that children from both groups were able to discriminate between sequential and random trials. DCD presents reduced procedural learning skills after controlling for global performance. This finding highlights the importance of considering the general functioning of the child while assessing learning skills in patients.

Developmental Coordination Disorder (DCD) affects 5–6% of school-aged children (American Psychiatric Association, 2013). This neurodevelopmental disorder is characterized by a reduced development of coordinated motor skills compared with age-matched peers despite opportunities to learn. These motor difficulties interfere with the child’s daily living activities and academic production. For example, children with DCD are often slower, more variable, and less accurate in riding a bike, handwriting, or...
performing self-care tasks. The onset of the symptoms occurs in the early developmental period and in the absence of any other medical condition (e.g., neurological condition affecting movements).

These clinical signs might relate to a lack of motor sequences automatization and suggest a deficit in procedural learning in DCD (Gheysen et al., 2011). Accordingly, procedural learning is the process enabling the acquisition and the progressive automatization of new perceptuomotor, perceptual, or cognitive skills through repetition and training (Cohen & Squire, 1980; Willingham, 1998). Reduced procedural learning may also account for a variety of motor or cognitive deficits frequently observed in DCD as in several other neurodevelopmental disorders (Nicolson & Fawcett, 2007), such as Developmental Language Disorder (DLD; e.g., Desmottes et al., 2016; Lum et al., 2014) or specific learning disabilities with impairment in reading (i.e., Specific Learning Disorder (SLD) with impairment in reading; e.g., Vicari (2005); Lum et al. (2013).

One typical paradigm used to assess procedural learning skills is the Serial Reaction Time task (SRTT) in which participants learn a visuomotor sequence through training (Nissen & Bullemer, 1987). In the SRTT, subjects have to answer as quickly and as accurately as possible to a stimulus by pressing a key corresponding to the stimulus location. Unbeknownst to the participant, a sequence is repeating over trials. As participants incidentally learn the visuomotor regularities, reaction times (RTs) for stimuli following the sequence (i.e., sequential trials) tend to diminish, while those for random trials (i.e., not following the sequence) elicit an abrupt increase of RTs (e.g., Destrebecqz & Cleeremans, 2001; Gheysen et al., 2011; Lejeune et al., 2013). The SRTT is commonly used to assess procedural learning skills as it allows to separately investigate two critical aspects, namely, (i) sequential learning and (ii) general motor learning (i.e., speed and visuomotor coordination) skills (Nissen & Bullemer, 1987), especially with paradigms alternating sequential and random conditions (Janacsek & Nemeth, 2012; Juhasz et al., 2019). Importantly, SRTT alternating between sequential and random conditions is particularly interesting as they enable to continuously assess sequential and general motor learning skills through training (Desmottes et al., 2016).

The few behavioral studies that investigated sequential procedural learning skills in DCD led to divergent results. In the seminal study of Wilson et al. (2003), no difference between children with and without DCD was observed using a classical SRTT performed with participant’s dominant hand. More specifically, in that study, the first four blocks were composed with stimuli following a determined (fixed) sequence, while the fifth block (i.e., transfer block) was composed with a random succession of stimuli. Results showed that children with severe to moderate DCD (Movement Assessment Battery for Children, M-ABC, score below the 15th percentile) were globally slower than typically developing (TD) peers to perform the five blocks (i.e., four sequential blocks and one transfer block). However, in both groups, the presentation of the transfer block elicited a similar increase of RTs, suggesting preserved procedural learning skills in DCD. Later, Gheysen et al. (2011) observed a specific alteration of sequential learning performance in children with severe DCD (Movement Assessment Battery for Children – Second Edition, MABC-2, score below the 5th percentile) using a SRTT performed bimanually, while global visuomotor habituation skills were preserved in DCD (after controlling for sample size, fatigue effect, and response frequency matching between random and sequential blocks). More precisely, RTs for sequential
trials decreased similarly through the task in both groups but, contrary to their TD peers, children with DCD were less able to differentiate random and sequential trials in the last blocks of practice. Inversely, using a touchscreen-based unimanual SRTT, which minimized task-related motor and cognitive constraints, Lejeune et al. (2013) reported similar sequence (procedural) learning performance between children with severe to moderate DCD and TD peers, suggesting that motor or cognitive confounds may have accounted for the sequential learning results that had been previously reported in DCD using classical SRTT (Gheysen et al., 2011). Of notice, in the two latter studies, similar signs of sequence awareness were reported between children with DCD and TD peers through the assessment of sequence awareness using a self-generation task (Gheysen et al., 2011; Lejeune et al., 2013).

Despite these discrepant behavioral results, strong neurophysiological evidence supports the procedural learning deficit hypothesis in DCD (Bo & Lee, 2013) as functional atypicalities (i.e., differing from the typical development) have been frequently reported within procedural learning-related brain networks in this disorder (Biotteau et al., 2016). For instance, reduced or enhanced functional brain activity (Debrabant et al., 2013; Reynolds et al., 2019; Zwicker et al., 2010, 2011) and connectivity (Cignetti et al., 2020; McLeod et al., 2014; Querne et al., 2008) have been observed in DCD as compared with TD peers in cortico-cerebellar and cortico-striatal networks that have been associated with procedural learning skills (Doyon et al., 2003; Janacsek et al., 2020). Moreover, basal ganglia and cerebellar dysfunctions have often been related to the DCD pathophysiology given their functional implication in motor learning/control and motor coordination/adaptation skills (Bo & Lee, 2013; Zwicker et al., 2009).

In parallel, executive functions, such as inhibition or working memory, which also rely on basal ganglia- or cerebellar-related cortico-subcortical networks (Niendam et al., 2012) are impaired in 40 to 60% of children and adults with DCD as compared to their TD peers (Alloway et al., 2009; Bernardi et al., 2016; Leonard et al., 2015; Wilson et al., 2017, 2020). The partial overlap between the cortico-subcortical networks underlying sequence learning skills and executive functions have led to hypothesize a possible association between these skills. Accordingly, it has been suggested that a lack of inhibition observed during a perceptual-motor learning task could reflect a learning deficit in DCD (Blais et al., 2017). In addition, early theoretical models of learning (e.g., the Adaptive Control of thought, ACT, Anderson, 1992) have suggested that learning evolves through different stages. First, a cognitive stage, where learning would rely on cognitive resources, such as working memory (Kim et al., 2013). Then, an associative stage (declarative and procedural) in which the involvement of cognitive skills reduces considerably and, finally, an automated stage (procedural) characterized by the involvement of psychomotor skills and perceptual speed. Despite inconsistencies across the literature regarding the links between working memory and incidental sequence learning performance (e.g., Jongbloed-Pereboom et al., 2019; Unsworth & Engle, 2005), some authors showed that stronger working memory skills predict better sequence learning performance (Bo et al., 2011; Martini et al., 2013). Moreover, according to the procedural deficit hypothesis developed in the context of DLD (Ullman & Pierpont, 2005) or SLD with impairment in reading (Krishnan et al., 2016), reduced sequential learning skills would be associated to a reduction of some oral or written language skills (mainly related to
phonology and syntax performance) and verbal working memory difficulties in these patients (see Ullman & Pierpont, 2005; Ullman et al., 2020).

Although previous behavioral studies investigating procedural learning skills in DCD have controlled for several confound factors (such as a possible fatigue effect or cognitive and/or visuomotor discrepancies between participants), additional confounds may have prevented a proper comparison of procedural learning performance between DCD and TD peers. In particular, SRTT studies classically measure the performance of participants through raw RTs. Yet, raw RTs might not be appropriate to compare groups with different RTs (Lukács & Kemény, 2015). Such discrepancies in RTs at baseline can be expected between children with DCD and their TD peers as motor slowness is one of the core symptoms of the disorder (American Psychiatric Association, 2013). Beside this motor slowness, children with DCD also frequently (in around 50% of the cases) present associated attention-deficit/hyperactivity disorder (ADHD), even in a subthreshold form (Kadesjö & Gillberg, 1998). Accordingly, evidence supporting a dimensional perspective of the disorder suggests that ADHD, characterized by symptoms of inattention, hyperactivity, or impulsivity (American Psychiatric Association, 2013), is not a discrete category but rather a continuous phenotype (for reviews, see Balázs & Keresztény, 2014; Drechsler et al., 2020). Still, subthreshold forms of ADHD, so as the formal diagnosis of ADHD, have an impact on the child daily life with functional deficits, psychosocial problems, and negative outcomes, and are frequently associated with other comorbidities (Balázs & Keresztény, 2014; Biederman et al., 2018; Norén Selinus et al., 2016).

Slower and less accurate responses due to inattention or impulsivity are also frequently reported in ADHD (Karatekin et al., 2009; Takács et al., 2017) strengthening the importance of controlling for global (motor) performance while assessing procedural learning skills in DCD. One solution to control for these aspects and, in particular, for baseline RTs differences and intra-individual variability, is to convert the child’s RTs into normalized RTs based on their mean or median RT (Lukács & Kemény, 2015; Lum et al., 2012; Thomas et al., 2004). Controlling for commission errors might also be highly relevant. Indeed, speed is the performance index classically used to investigate sequence learning skills, regardless of commission errors (Laventure et al., 2016), considering that the error rate is quite low in the general population (e.g., Meulemans et al., 1998). However, even small differences in accuracy can reflect that participants adopt different motor strategies (i.e., speed-accuracy trade-off; Laventure et al., 2016; Liesefeld & Janczyk, 2019). Hence, a better score can result from a higher speed (i.e., slower RTs) or a better accuracy (i.e., lower error rate; Vandierendonck, 2021). It has been suggested that children with DCD might favor accuracy over speed (Farmer et al., 2016). Yet, the speed-accuracy trade-off can vary across and within participants (Liesefeld & Janczyk, 2019), strengthening the relevance of investigating sequence learning using appropriate measures that combined both properties of motor performance (i.e., speed and accuracy). To date, combined measures of speed and accuracy have never been used to characterize the sequence learning skills of children with DCD, neither normalized RTs were used to control for baseline RTs differences and intra-individual variability.

Yet, the neurophysiological evidence surmising atypical procedural learning-related brain processes in DCD (e.g., Biotteau et al., 2016; Bo & Lee, 2013; Cignetti et al., 2020) and the discrepant behavioral results reported in the literature (Gheysen et al., 2011; Lejeune et al., 2013; Wilson et al., 2003) call for a better understanding of procedural
learning skills in DCD. To fill this gap, we investigated procedural learning skills using a touchscreen-based SRTT with a repeating sequence alternating with random successions of trials in a group of children with DCD and age-matched TD peers. Hence, we hypothesized that controlling for global performance through normalized RTs and using combined measures of speed and accuracy will help to better characterize procedural learning difficulties in DCD. According to the procedural learning deficit hypothesis in DCD (Gheysen et al., 2011; Nicolson & Fawcett, 2007), we expected to find a weaker ability to learn new visuomotor sequences in children with DCD as compared to TD peers, possibly reflected by a lower evolution of their sequence learning performance through the SRTT training. We also expected that children with and without DCD would present similar levels of sequence awareness (Gheysen et al., 2011; Lejeune et al., 2013). Finally, given the partial overlap between brain circuits involved in procedural learning (Doyon et al., 2003; Janacsek et al., 2020) and high-level cognitive functions (Niendam et al., 2012), we also expected to find some associations between sequential learning skills, inhibition, and verbal working memory skills in our participants. As the severity of DCD symptoms might possibly explain discrepancies between previous SRTT studies (i.e., reduced procedural sequence learning skills in children presenting a severe DCD with an MABC-2 score below the 5th percentile and preserved procedural learning skills in children with DCD with an MABC-2 below the 15th percentile; Gheysen et al., 2011; Lejeune et al., 2013; Wilson et al., 2003), we also expected to find an association between sequence learning performance and motor coordination.

Methods

Participants

Seventy-four children with and without DCD aged between 7 and 11 years old were recruited to take part in the study. Descriptive information regarding the two groups is presented in Table 1. Ten participants were excluded as they did not fulfill the following inclusion criteria or met an exclusion criterion (6 DCD and 4 TD), one child due to a technical issue with the software of the SRTT (1 DCD) and one child failed to complete the task due to excessive motor agitation (1 DCD). Final samples were therefore composed of 31 children with DCD (3 females and 28 males, mean age ± SD: 9.61 ± 1.52 years) and 31 TD children (13 females and 18 males, mean age ± SD: 9.83 ± 1.37 years).

Inclusion criteria for the children with DCD were the four diagnosis criteria of the DSM-5 (American Psychiatric Association, 2013). These criteria were assessed as follows: (i) the global score at the French version of the MABC-2 (measure of the motor skills; Henderson et al., 2007; Marquet-Doléac et al., 2016) is equal or below the percentile 15th (two children were excluded as they scored at the percentile 16th), (ii) the score at the Developmental Coordination Disorder Questionnaire (DCD-Q; Martini et al., 2011), a parental questionnaire measuring the impact of motor difficulties on the child’s life, is in the suspected or indicative range of DCD, (iii) a short anamnesis with at least one parent reports the onset of the symptoms in the early developmental period, and (iv) the presence of neurological disorder is address through the short anamnesis and any intellectual disability with the verbal comprehension index of the WISC-V (Wechsler, 2016). Children with DCD
presented severe (n = 21; MABC-2%ile ≤ 5th) to moderate (n = 10; percentile 6–15th) motor impairments.

Children with TD were included if they did not present any history of motor difficulties, their MABC-2 global score was equal or above the percentile 25th (four children were excluded as they scored below the percentile 25th), the DCD-Q was not indicative of a (suspected) DCD, and they did not meet any of the following exclusion criteria. Moreover, children from both groups had to present normal to corrected vision. Exclusion criteria were the same for both groups: intellectual disability (with the verbal comprehension index of the WISC-V < 80; four children from the DCD group were excluded based on this criterion), autism spectrum disorder, history of psychiatric or neurological disorder, or born very preterm (<33 weeks gestational age; all controlled through a short anamnesis). In addition, TD children should not present any neurodevelopmental disability (controlled with the short anamnesis and the parental questionnaire ADHD-RS-IV, score >28 is indicative of ADHD; DuPaul et al., 1998).

In children with DCD, concurrent ADHD diagnosis was not an exclusion criterion (n = 19), as sequence-specific aspects seem preserved in the ADHD population (Sanjeevan et al., 2020; Takács et al., 2017). Associated symptoms might have a deleterious effect on the general aspects of the task (i.e., RTs and accuracy; Karatekin et al., 2009; Staels & Van den Broeck, 2017; Takács et al., 2017) and were controlled by converting RTs into normalized RTs. ADHD was assessed according to DSM-5 criteria (American Psychiatric Association, 2013) by a multidisciplinary team including pediatric neurologists and neuropsychologists. Methylphenidate medication for ADHD (n = 10) was interrupted at least 24 h before the experiment. Descriptive information regarding children with and without associated diagnosis of ADHD can be found in supplemental material 1. The ADHD comorbidity and medication intake were characterized in children with severe (children with DCD+ADHD: 13/21; methylphenidate medication: 6/21)

Table 1. Descriptive information.

<table>
<thead>
<tr>
<th></th>
<th>DCD (n=31)</th>
<th>TD (n=31)</th>
<th>Statistics</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex, n F/M</td>
<td>3/28</td>
<td>13/18</td>
<td>X²(62) = 8.42</td>
<td>&lt;.01</td>
</tr>
<tr>
<td>Latency, n R/L/A</td>
<td>26/4/1</td>
<td>29/2/0</td>
<td>X²(62) = 1.83</td>
<td>.40</td>
</tr>
<tr>
<td>Age (years)</td>
<td>9.61 (1.52)</td>
<td>9.83 (1.37)</td>
<td>t (60) = -0.61</td>
<td>.55</td>
</tr>
<tr>
<td>DCD-Q</td>
<td>33.63 (10.96)</td>
<td>65.29 (6.06)</td>
<td>t (45) = -13.90a</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>ADHD-RS-IV score</td>
<td>31.13 (12.18)</td>
<td>11.74 (7.50)</td>
<td>t (50) = 7.55a</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Pathological ADHD-RS-IV, n yes/no</td>
<td>20/11</td>
<td>0/31</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>MABC-2 (pc)</td>
<td>3.67 (3.98)</td>
<td>50.71 (21.63)</td>
<td>t (32) = -11.90a</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Manual Dexterity (SN)</td>
<td>3.77 (1.63)</td>
<td>9.45 (2.66)</td>
<td>t (50) = -10.15a</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Aiming and catching (SN)</td>
<td>7.00 (2.28)</td>
<td>10.16 (2.44)</td>
<td>t (60) = -5.27</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Static-dynamic balance (SN)</td>
<td>5.61 (3.31)</td>
<td>11.10 (1.62)</td>
<td>t (44) = -8.28a</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Verbal Comprehension Index (WISC-V)</td>
<td>105.45 (16.90)</td>
<td>114.32 (12.83)</td>
<td>t (60) = -2.33</td>
<td>.02</td>
</tr>
<tr>
<td>Go-NoGo (commission errors, z-score)</td>
<td>-1.29 (1.02)</td>
<td>-0.84 (1.02)</td>
<td>U = 337 b</td>
<td>&lt;.04</td>
</tr>
<tr>
<td>Backward digit span (z-score)</td>
<td>-0.41 (1.55)</td>
<td>0.73 (1.05)</td>
<td>U = 231 b</td>
<td>&lt;.001</td>
</tr>
</tbody>
</table>

Values are presented as mean (standard deviation), except for sex, laterality and pathological ADHD-RS-IV score. = Developmental Coordination Disorder; TD = Typically Developing children; F = female, M = Male; Laterality = Edinburgh Handedness Inventory (Oldfield, 1971), R = right-handed, L = left-handed, A = ambidextrous; DCD-Q = The Developmental Coordination Disorder Questionnaire (Martini et al., 2011); ADHD-RS-IV = Attentional Deficit Hyperactivity Disorder Rating Scale IV (DuPaul et al., 1998); MABC-2 = Movement Assessment Battery for Children, 2nd ed. (Marquet-Doléac et al., 2016), percentile (pc) of the general scale and standard notes (SN) of the 3 subscales are reported; WISC-V = Wechsler Intelligence Scale for Children, 5th ed. (Wechsler, 2016).

Nonparametric Welch’s t test, in cases of violation of homogeneity of variance. b Nonparametric Mann–Whitney test, in cases of violation of normality.
or moderate DCD (children with DCD+ADHD: 6/10; methylphenidate medication: 4/10). In all participants, the severity of ADHD comorbidity was assessed through the ADHD-RS-IV (DuPaul et al., 1998) parental questionnaire (mean ± SD : DCD with ADHD = 36.52 ± 9.91, DCD without ADHD = 22.58 ± 10.66), one of the most used DSM-based questionnaires assessing the main symptoms of inattention and hyperactivity/impulsivity validated in multiple languages (Döpfner et al., 2006). Participants were screened for other neurodevelopmental disorders through a short parental anamnesis and available medical files, given the high prevalence of associated disorders in DCD (Dewey, 2018; Visser, 2003). Ten children with DCD and associated ADHD also presented other associated comorbidities (five presented one additional disorder and five multiple disorders). Six children presented an associated SLD with impairment in reading, 3 in writing, 5 in arithmetic, and 3 an associated DLD.

Children from the DCD group were recruited through neuropediatricians and health professionals’ consultations (n = 20) as well as social medias (n = 9) and public primary schools (n = 2). Children from the TD group were recruited through acquaintances (n = 20), social medias (n = 7), and in primary schools after receiving the approval of the authorities (n = 4), in the French-speaking part of Belgium. Written informed consents were obtained, after approval of local Ethics Committee (CUB Hôpital Erasme and Hôpital Universitaire des Enfants Reine Fabiola; Brussels, Belgium).

**Material and procedure**

The Serial Reaction Time task (SRTT) used to assess procedural learning was presented as a cars race game (based on Cars Cartoons© and designed by Desmottes et al., 2016, see Figure 1). A different car was presented in each block in order to maintain the

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**Figure 1.** Experimental design. **Top.** Motor and neuropsychological assessments. Motor skills (MABC-2 (Marquet-Doléac et al., 2016)), intellectual functions (verbal comprehension index, WISC-V (Wechsler, 2016)), motor inhibition (Go-NoGo task), visuospatial and verbal short-term memory (block tapping test; forward digit span), working memory (backward digit span). Experimental task. The serial reaction time task (SRTT, designed by (Desmottes et al., 2016)) lasted for approximately 30 min and was composed of 10 blocks of practice including intermixed sequential (gray strips) and random (white strips) successions of trials. In further analyses, consecutive blocks were averaged in 5 epochs of 2 consecutive blocks. In each trial, the stimulus appeared in one of the four quadrants of the touchscreen (Bottom), numbered from 1 to 4 (numbers and white dashed lines were not displayed during the task). Self-generation task. Following the SRTT, children were asked to generate what they remembered from the repeating sequence on the same display.
child’s motivation and as new stimulation improves attentional vigilance (Baijot et al., 2016; Zentall & Zentall, 1983). Children could take short breaks between the blocks when needed. They were then encouraged to keep motivation in winning the race and to start the next block as soon as they were feeling ready. Participants were asked to touch as quickly and accurately as possible a stimulus appearing in one quadrant of a touchscreen with their dominant hand (unimanual). This version of the SRTT has been widely used to investigate sequential learning skills in children with learning disabilities (e.g., DLD; Desmottes et al., 2016, 2017; Gabriel et al., 2012, 2013), due to its suitability to test the procedural deficit hypothesis in children with neurodevelopmental disorders (i.e., game presentation, reduction of fine motor and cognitive constrains). Quadrant presentation was used instead of linear presentation as it enables to separate the locations into large spatial domains (Lejeune et al., 2013; Thomas & Nelson, 2001). Participants were not informed of the presence of a repeating sequence. The stimulus disappeared after touching any location of the screen (250 ms response-stimulus interval) or in the absence of response (after 4300 ms). The screen was divided into four quadrants and the sequence comprised 10 locations of stimuli (2-4-1-3-4-2-1-4-3-1, see locations in bottom part of Figure 1). Each location appeared in a similar proportion during the random and sequential successions of trials (1–4 = 30%; 2–3 = 20%). The learning session included 10 training blocks of 84 stimuli. As first developed by Meulemans et al. (1998), each of the 10 training blocks started with 4 random stimuli (considered as training stimuli that were not further used in the analyses), followed by 10 sequential stimuli and then by 6 random stimuli, and so on. A training block included five presentations of the repeating sequence (10 stimuli), alternating with random successions of stimuli (6 stimuli), allowing for the measurement of sequence learning skills continuously. The E-Prime software (version 2.1) was used for stimulus presentation and data recording.

As in Desmottes and colleagues’ study (Desmottes et al., 2016), sequence awareness was assessed through a qualitative interview including a series of specific questions, and a self-generation task during which children were asked to spontaneously produce the sequence on the touchscreen as during the SRTT practice. Interviews occurred as followed: (i) “Did you notice anything in the game?,” (ii) “Did you observed that the cars followed sometimes a sequence?,” and (iii) “Did you use this observation to try to move faster or commit less errors?.” Children were then informed that a sequence was determining cars locations: “The car location followed a fixed sequence of positions on the screen boxes. Can you describe any pattern you think might have been there?.” They were asked to try reproducing the sequence (they thought they had learned) on the touchscreen during 30 trials (i.e., 30 touch on the screen; 3 repetitions of the 10-item sequence). They were encouraged to “guess the sequence” if they thought not being able to recall any regularity. Generation score (Desmottes et al., 2016; Gheysen et al., 2011; Lejeune et al., 2013) was calculated as the number of correct triplets (maximum = 28) with chance level set at 6.22 (see Desmottes et al., 2016 for details regarding methods and analyzes).

Additional cognitive functions were assessed in our participants, either on the same day (alternating the order morning-afternoon between the cognitive tests and the procedural SRTT practice between participants; TD: n = 24; DCD: n = 2) or during a second day to avoid fatigue effect (alternating the order day1-day2; TD: n = 7; DCD: n = 29).
Motor inhibition was assessed using a Go-NoGo task (Zimmermann & Fimm, 2004) where children responded as quickly as possible to a target “x” (50% of trials) but not to a distractor “+.” Number of commission errors (response to a distractor) were counted. Backward digit span test (WISC-V; Wechsler, 2016) was used to assess verbal working memory: participants had to recall growing sequences of digits in backward order. Final scores corresponded to the higher sequence of digits repeated successfully. To control for age differences within the sample, the raw values have been standardized (z-transformed) based on the mean and the standard deviation of the normative data for each child.

**Statistical analyses**

Demographic data, motor, and neuropsychological assessment scores were compared between children with DCD and TD children using chi-square tests for categorical variables, and two-tailed unpaired t tests for continuous variables. These results are reported in Table 1. Nonparametric Welch’s t tests were used in cases of violation of variance homogeneity and a Mann–Whitney tests in cases of normality violation.

Two specific learning indices were computed based on SRTT results: a sequence-specific learning index (SSLI) and a global performance index (GPI). Indices measured on the 10 training blocks of the SRTT were averaged to be analyzed into 5 epochs of 2 consecutive blocks (see Figure 1) as in previous study (Desmottes et al., 2016). The term “Epochs” is used instead of the traditional “Blocks” to avoid confusion between the blocks (i.e., training blocks while the child performs the task) and the epochs (i.e., used in further analyses).

The SSLI measure was based on z-transformed RTs, which has the advantage to control for baseline RTs difference between groups (Lukács & Kemény, 2015) and within-subject motor speed variability (Lum et al., 2012). Baseline RTs differences can be expected between children with DCD and TD peers as motor slowness and variability have consistently been described in this disorder (e.g., Farmer et al., 2016; Missiuna et al., 2003). Accordingly, between-group baseline difference has been observed (see results section: Serial reaction time task) when comparing the median RTs in Epoch 1 between children with DCD and TD peers, using an unpaired t test (or a nonparametric equivalent in case of violation of any assumption).

**Before calculating the z-transformed RTs,** we excluded for each subject the errors (and their following n + 1 trial, as post-error slowing is frequently observed; Ceccarini & Castiello, 2018), omissions and correct trials associated with (i) RTs above the mean +3SD for each training block and condition (considered as potential attention lapses; Borragán et al., 2016; Lum et al., 2012), or (ii) RTs <100 ms (considered as anticipations; Debrabant et al., 2013). An overall median RT and associated overall standard deviation were then computed for each participant across all epochs and conditions. For each trial, a z-transformed individual RT was then computed by subtracting the participant’s overall median performance (see above) from each raw RT and dividing this value by the participant’s overall standard deviation (Lukács & Kemény, 2015; Lum et al., 2012; Thomas et al., 2004). Z-transformed RTs were then averaged by condition and training block for each participant. The SSLI was then measured as a difference between the averaged z-transformed RTs in the random and the sequential conditions (“Random – Sequential;” Thomas et al., 2004) for each participant and training block, and then
averaged by epoch. This SSLI has the advantage to assess sequence-specific learning throughout the task, independently of task-general skills (e.g., visuomotor coordination, improvement of speed processing; Juhasz et al., 2019).

An increase of SSLI values over epochs reflects sequence learning through training with greater differences of RTs between random and sequential trials. A repeated measures ANOVA was conducted on the SSLI with 1 within-subjects factor (Epoch: 1–5), and 1 between-subjects factor (Group: DCD vs. TD).

Procedural learning was also assessed through a speed-accuracy trade-off measure (Liesefeld & Janczyk, 2019), namely the Global Performance Index (GPI, adapted from Laventure et al., 2016) computed by combining speed (i.e., the gold-standard performance index in SRTT studies) and accuracy measures for each participant. To this end, for each participant, we first computed a “speed index” based on z-scored RTs referenced to the median RTs (and SD) across all trials (i.e., including correct trials and errors, only the omissions were removed from this index) to control for intra-individual variability. Then, an “accuracy index” was computed as the number of commission errors per number of responses. Using these measures, we computed a GPI, assessing the speed/accuracy trade-off, for each training block and condition, and then averaged by epoch, as:

\[
GPI = e^{-\text{speed}} * e^{-\text{accuracy}}.
\]

“e” represents the Euler’s number, a mathematical constant defined as the base of the natural logarithm (~2.71828). A higher GPI value reflects a better performance in speed or accuracy. A second repeated measures ANOVA was thus computed on the GPI measures with 2 within-subjects factors (Epoch: 1–5; Condition: Sequential vs. Random) and 1 between-subjects factor (Group: DCD vs. TD).

To assess the critical use of normalized indices to control for baseline RTs differences between groups, we also performed a third repeated measures ANOVA on raw RTs performance (i.e., often used in SRTT studies) with 2 within-subjects factors (Epoch: 1–5; Condition: Sequential vs. Random) and 1 between-subjects factor (Group: DCD vs. TD). As for the SSLI analyses, errors trials and their following n + 1 trial, omissions, and correct trials with RTs above the mean +3SD or <100 ms were discarded from the raw RTs analysis.

As children with comorbid ADHD were included in our study, our results were controlled for the impact of the ADHD comorbidity severity. To do so, we performed a repeated-measures ANCOVA on the two learning indices (SSLI and GPI) with the same within- and between-subject factors, and the score at the parental questionnaire ADHD-RS-IV (centered; Delaney & Maxwell, 1981; DuPaul et al., 1998) as a covariate.

For each ANOVA, the normality of the data (Shapiro-Wilk’s test), the homogeneity of variance (Levene’s test), and the sphericity (Mauchly’s test) were verified. When sphericity could not be assumed, results of the Greenhouse-Geisser were reported. The homogeneity of the regression slopes (non-significance of the interaction between the covariate and the between-subject variable; Field, 2017) was verified for the ANCOVAs.

We computed correlational analyses on the complete sample of children to test a priori associations between specific cognitive functions (i.e., motor inhibition and verbal working memory) and the individual linear slope of SSLI. To control for age differences within the sample, the raw values have been standardized (z-transformed) based on the mean...
and the standard deviation of the normative data for each child. Furthermore, the
association between sequence learning performance (i.e., SSLI slope) and motor coordi-
nation (i.e., global MABC-2 score) was also assessed through a correlational analysis.
When normality could not be assumed or the data included outliers, a correlation
coefficient applied to ranked data, the Spearman’s rho, was used instead of a Pearson’s
correlation analysis (Field, 2017). Participants were considered as outliers when they
were deviating from more than 3 median absolute deviation from their group for each
behavioral measure (Leys et al., 2013).

To assess sequence awareness, the generation scores were compared to the chance
level (set at 6.22) using a one-sample t test (or a nonparametric Wilcoxon rank-sum test
in cases of violation of the normality). The generation scores of the two groups were also
compared using an independent t test (or a nonparametric Mann–Whitney test in case of
violation of normality).

Finally, complementary analyses were conducted on the two learning indices (SSLI
and GPI) (i) while controlling for sex and verbal comprehension index to rule out
a potential effect of these confound factors on our experimental results, (ii) on right-
handed children only, (iii) and by comparing children with DCD with and without
methylphenidate medication. Complementary analyses were also conducted on the
percentage of errors. These analyses are presented in supplemental material 2–6.

Statistical analyses were performed on R Studio (RStudio Team, 2015) and on
JAMOVI Software (The jamovi project, 2021).

Results

Serial reaction time task

Before calculating the SSLI, several trials were excluded for each subject: errors and their
following trial (mean ± SD [range] in %: DCD = 9.52 ± 9.25 [0.5–35.8]; TD = 4.46 ± 5.09
[0.5–20.5]), omissions (DCD = 0.17 ± 0.42 [0–1.63]; TD = 0.05 ± 0.21 [0–1.13]), correct
trials above 3SD from the mean for each epoch and condition (DCD = 1.84 ± 0.48 [1–
2.88]; TD = 2.26 ± 0.5 [0.87–3.13]), and with RTs <100 ms (DCD = 0.03 ± 0.08 [0–0.37];
TD = 0.01 ± 0.07 [0–0.37]). Baseline RTs (i.e., median RTs in Epoch 1) were compared
between the groups and showed that children with DCD (mean ± SD: 733 ± 86.4 ms)
were slower than their TD peers (689 ± 69.8 ms; U = 324; p = .03).

The repeated measures ANOVA on the SSLI revealed a main effect of the Epoch (F
(4,240) = 10.53; p < .001; ηp²= .15), a main effect of the Group (F(1,60) = 5.41; p = .02;
ηp²= .08), but no interaction between these factors (p=.67). As a reminder, unlike the
other indices, the factor Condition (sequential vs. random) was not included in the
SSLI-related statistical analyses as this index was measured as a difference between the
averaged z-transformed RTs in the random and the sequential conditions, enabling to
assess sequence-specific learning performance throughout the task. The assumptions
of normality, homogeneity of variances and sphericity were verified. Tukey’s post-hoc
analyses revealed significant improvement between the first epoch and all the sub-
sequent epochs (ps < .02, all other ps >.14). Importantly, to control for the severity of
the ADHD comorbidity, a repeated measures ANCOVA was computed on the SSLI
and revealed similar results, with a main effect of the Epoch (F(4,236) = 10.45; p
<.001; η² = .15) and the Group (F(1,59) = 6.03; p = .02; η² = .09), but no interaction between these factors (p = .62). Figure 2(a) shows the performance of the two groups on the SSLI through the 5 Epochs of the SRTT training. Hence, these results showed an overall reduced sequence learning performance in children with DCD as compared with matched TD (see Figure 2b).

The repeated measures ANOVA conducted on GPI revealed a main effect of the Epoch (F(3.27, 195.97) = 12.62; p < .001; η² = .17), the Condition (F(1,60) = 110.24; p < .001; η² = .65), and the Group (F(1,60) = 11.5; p = .001; η² = .16). Significant Epoch × Condition (F(4,240) = 10.79; p < .001; η² = .15), Epoch × Group (F(3.27,195.97) = 3.58; p = .007; η² = .06), and Condition × Group (F(1,60) = 7.21; p = .009; η² = .11) interactions were also reported, while the interaction Epoch × Condition × Group was not significant (p = .91). As sphericity could not be assumed for the Epoch, Greenhouse–Geisser results are reported. The assumptions of normality and homogeneity of

![Figure 2](image.png)

**Figure 2.** a. Evolution of Sequence-Specific Learning Index (SSLI) across epochs by group: Developmental Coordination Disorder (DCD) and Typically Developing (TD) children; with a significant main effect of the Epoch and the Group (non-significant interaction). b. Significant main effect of the Group: differences of SSLI overall mean between groups. c. Evolution of Global Performance Index (GPI) across epochs by condition for each group (DCD and TD), with significant main effects of the Epoch, Condition, Group and significant Epoch × Condition, Epoch × Group, and Condition × Group interactions (non-significant Epoch × Condition × Group interaction). d. Significant interaction between Group and Condition factors on GPI. Tukey post-hoc analyzes showed that both groups presented better performance for sequential trials compared with random successions of trials, with a significant difference between the groups in the sequential condition. Error bars represent 95% of the confidence intervals. * p < .05.
variances were verified. Tukey post-hoc analyses demonstrated that children with DCD (mean GPI difference = 0.10; t(60) = 5.53; \( p < .001 \); \( d = 1.07 \)) as well as TD peers (mean GPI difference = 0.17; t(60) = 9.32; \( p < .001 \); \( d = 1.57 \)) showed an overall better GPI for sequential compared to random trials. Yet, a significant difference was found between the groups in sequential (t(60)=−3.39; \( p = .007 \); \( d = −0.86 \)), but not in random trials (t(60)=−1.24; \( p = .60 \); \( d = −0.31 \)), showing a higher discrimination between sequential and random trials in TD children (see Figure 2d). After controlling for the severity of the ADHD comorbidity, the repeated measures ANCOVA computed on the GPI revealed similar results, with main effects of the Epoch (F(3.24, 191.02) = 12.60; \( p < .001 \); \( \eta^2_p =.18 \)), Condition (F(1,59) = 110.34; \( p < .001 \); \( \eta^2_p =.65 \)) and Group (F(1,59)=.58; \( p = .008 \); \( \eta^2_p =.11 \)), and significant Epoch \( \times \) Condition (F(4,236) = 10.79; \( p < .001 \); \( \eta^2_p =.15 \)) and Condition \( \times \) Group (F(1,59) = 6.98; \( p = .01 \); \( \eta^2_p =.11 \)) interactions. However, the interaction Epoch \( \times \) Group was no longer significant (all other \( ps > .18 \)).

To assess the added value of using normalized indices, complementary repeated measures ANOVA analyses were performed on the raw RTs with two within-subject factors (Epoch: 1–5, Condition: sequential vs. random) and one between-subject factor (Group: DCD vs. TD). The ANOVA analysis revealed a significant effect of the Epoch (F(2.56,153.64) = 11.28; \( p < .001 \); \( \eta^2_p =.16 \)), the Condition (F(1,60) = 203.39; \( p < .001 \); \( \eta^2_p =.77 \)), the Group (F(1,60) = 10.5; \( p = .002 \); \( \eta^2_p =.15 \)), and a significant interaction between Epoch \( \times \) Condition (F(4,240) = 9.92; \( p < .001 \); \( \eta^2_p =.14 \); all other \( ps > .06 \)). The effect of the group was no longer significant (\( p = .26 \)), suggesting that comorbid ADHD impacted the slower RTs of children with DCD.

We conducted complementary analyses to rule out a potential effect of confound factors (e.g., effect of the sex or the Verbal Comprehension Index on the learning performance) and the heterogeneity of the children (i.e., laterality of the children and methylphenidate medication intake in the DCD group) on the experimental results. Complementary analyses were also conducted on the percentage of errors. These complementary analyses are presented in the supplemental material 2–6.

Complementary correlation analyses between procedural learning measure (SSLI improvement) and specific cognitive skills were performed based on a priori hypotheses. Results showed significant correlations between the SSLI slope and (a) motor inhibition (\( r_s = 0.36; p = .004 \)), (b) verbal working memory (\( r_s = 0.31; p = .01 \)), and (c) motor skills (\( r_s = 0.27; p = .03 \); see Figure 3 for details). As normality could not be assumed for the three measures and outliers were found in motor inhibition (n = 3) and verbal working memory (n = 1) data, a Spearman’s rho test was used for these analyses instead of Pearson’s correlation.
Sequence awareness

The qualitative analyses of interviews showed that only a few children spontaneously reported being aware of a repeating sequence (i.e., answering that they noticed a repetitive pattern to the first question; DCD: 5/31; TD: 3/31) while most of the children reported to be aware of this pattern when they were specifically asked if they had noticed regularities in the succession of car’s locations (i.e., answering “yes” to the second question; DCD: 22/31; TD: 24/31).

Yet, using the generation task, in which participants were asked to produce what they could remember from the sequence, Mann-Whitney test showed no significant group difference (p = .77) in sequence awareness. Generation scores of children with DCD (mean ± SD: 7.42 ± 3.86; t(30) = 1.73; p = .09) and TD (7.63 ± 4.62; W = 270; p = .45) did not differ from chance level. When normality of the data could not be assumed, nonparametric analyses have been performed.

Discussion

This study aimed at testing the procedural learning deficit hypothesis in children with DCD while controlling for global performance aspects (i.e., differences in baseline RTs and intra-individual variability). Results highlighted poorer level of sequential learning performance in children with DCD, as shown by a global reduction of sequence-specific learning index (SSLI) compared to age-matched TD peers. Yet, the improvement of their performance through the training suggests that children with DCD had learned the repeating sequence, but to a lesser extent than their TD peers. Complementary analyses performed on global performance index (GPI, combining RTs and accuracy) revealed that children from both groups were able to discriminate sequential from random trials but with a greater mean difference between trial types for TD children than for children with DCD. In that same line, learning slope analyses showed that children with DCD were able to improve their performance through training to a similar extend than TD peers (i.e., no significant interaction between the Group × Epoch for the SSLI or Group ×
Epoch x Condition for the GPI). However, the sequence learning skills of children with DCD were globally reduced in comparison with TD (i.e., significant Group effect for the SSLI and Group x Condition for the GPI), reflecting a general lower ability to learn the sequence (i.e., a lower level of learning) as their performance remained reduced compared to TD peers throughout the training. Moreover, a complementary correlation analysis revealed that the more severe were motor coordination symptoms in DCD the lower were their ability to learn the sequence across epochs through the task. Altogether, this suggests that DCD is characterized by a lower ability to learn sequential regularities in their environment although the discrimination and the learning of a repeating sequence remains possible (as highlighted by GPI and learning slope analyses respectively).

Contrary to previous behavioral studies assessing procedural learning skills in DCD, we computed SSLI based on normalized RTs to measure the differences between random and sequential trials. It thus enabled us to control for baseline RTs differences between groups and to reflect a proportional magnitude of sequence (procedural) learning in our participants (Lukács & Kemény, 2015; Lum et al., 2012). On the other hand, another original aspect of our study is that we used GPI measures, combining both RTs and errors, and thus allowing to track participants different motor strategies (i.e., speed-accuracy trade-off; Laventure et al., 2016; Liesefeld & Janczyk, 2019) to assess procedural learning skills in DCD and TD peers. Combining RTs and accuracy through GPI measures was particularly relevant as children with DCD tend to be slower, more variable and inaccurate than their TD peers to perform motor actions (American Psychiatric Association, 2013). As expected, the results obtained using the sequence-specific learning index based on normalized RTs contrast with those that we obtained when the analyses were computed on raw RTs. Discrepancies between raw RTs and normalized learning index are common in the literature. For example, a recent meta-analysis investigating procedural learning skills during lifespan showed that the use of raw RTs or normalized indices leads to different results in terms of developmental trajectories (Zwart et al., 2019). While procedural learning analyses performed on a normalized index led to an inverted-U shape performance from childhood to late adulthood, with adults outperforming children and elderly, raw RTs-based analyses lead to better performance during childhood followed by a progressive decrease until late adulthood. Importantly, it has been argued that normalized data are more suitable to investigate age-related differences in procedural learning skills as raw RTs data may lead to groups comparisons with different baseline RTs (Lukács & Kemény, 2015). The use of a normalized index such as SSLI seems also more appropriate to assess sequence learning (and discrimination) skills in children with DCD, their baseline (motor) performance being globally slower than TD peers. Hence, our findings bring significant information to previous behavioral studies reporting discrepant results on sequence learning skills in DCD (Gheysen et al., 2011; Lejeune et al., 2013; Wilson et al., 2003). The specific learning indices used in our study may have played a key role in the clarification of sequence learning difficulties in children with DCD compared to previous studies.

Our results also highlighted an association between procedural learning skills and motor coordination performance. Although the slope of sequence learning performance did not significantly differ between groups, we observed that children with more severe DCD symptoms were also showing a reduced slope of sequence learning skills through
the task. The severity of the DCD symptoms (i.e., motor coordination) might then be an important factor to consider while investigating procedural learning skills in DCD and that may also explain the discrepancies between behavioral SRTT studies (Gheysen et al., 2011; Lejeune et al., 2013; Wilson et al., 2003). Our results further suggest that procedural learning deficits might be more prominent in a subset of children with DCD, as highlighted by complementary correlation analyses. These analyses showed significant associations between individual sequence learning skills through the task (i.e., linear slope of SSLI) and motor inhibition or verbal working memory skills. These correlations indicated that children who showed poorer sequence learning skills also showed weaker motor inhibition (positive correlation with the standardized errors in Go-NoGo task) and verbal working memory (positive correlation with the standardized digits sequence size in Backward Digit Span). These associations could be related to previous evidence indicating partly overlapping brain networks between these functions. Accordingly, cortico-striatal and cortico-cerebellar networks have been related to sequence learning (Doyon et al., 2003; Janacsek et al., 2020) but also to underlie executive functions (such as working memory and inhibition; Niendam et al., 2012). Therefore, weaker sequence learning skills in our participants could rely on an atypical functioning of brain networks (e.g., Biotteau et al., 2016; Bo & Lee, 2013; Cignetti et al., 2020; McLeod et al., 2014; Querne et al., 2008; Zwicker et al., 2010, 2011) also supporting the executive impairments often observed in DCD (Alloway et al., 2009; Bernardi et al., 2016; Leonard et al., 2015; Wilson et al., 2020). Considering the clinical heterogeneity of neurodevelopmental disorders, it can be hypothesized that lower sequential learning skills could be associated to a combination of motor, inhibitory and working memory skills impairments. Our results call for further investigations of the associations between specific cognitive-motor profiles and procedural learning skills in neurodevelopmental disorders.

Finally, in our study, very few children (a maximum of five children per group) spontaneously reported being aware of the presence of a repeating sequence in the task while answering the basic open question “Did you notice anything in the game?.” This result is in line with previous observations reported in the DCD (5 children per group, DCD or TD, Gheysen et al., 2011) or DLD populations (where none of the DLD or TD children reported an awareness of the sequence (Desmottes et al., 2016). Worth noticing, our study, on the other hand, highlighted that a large proportion of children report noticing the presence of a repeating sequence when they answered a second and more precise closed question (i.e., “did you notice the presence of a repeating pattern while the car was moving between boxes”). These discrepancies highlight the importance of the question form while investigating subjective sequence awareness in children. As expected, the degree of sequence awareness objectively assessed at the end of the SRTT practice with the generation task was similar between children with DCD and age-matched TD peers but, unlike previous studies (Gheysen et al., 2011; Lejeune et al., 2013), did not differ from chance level. The SRTT variant used in this study may explain this discrepancy as explicit knowledge may be more difficult to acquire in the context of a sequential learning task including alternating sequential and random series of trials (Mayor-Dubois et al., 2016). Of note, explicit knowledge, possibly resulting from representations of higher quality, is a reliable information to perform more efficiently the SRTT (Destrebecqz & Cleeremans, 2001; Stefaniak et al., 2008). The quality of the representations refers here as the strength, distinctiveness,
and stability in time of the memory traces (Cleeremans, 2004). Considering our results, differences in sequence awareness do not seem to explain reduced procedural learning skills in DCD as compared with TD. Other confound factors such as IQ-related between groups differences should not account for the observed results as tested through complementary analyses and as participants showed intellectual skills within normal range.

One limitation is the unequal sex ratio between our two groups. There is typically a higher rate of boys in DCD (between 2:1 and 7:1) and more broadly in neurodevelopmental disorders (American Psychiatric Association, 2013). Yet, complementary analyses suggested that the sex factor did not impact our results as procedural learning performance did not differ between TD girls and boys on one index, and boys (overrepresented in the DCD sample) outperformed girls on the second index. Another limitation relates to the fact that this study focused on the early stages of motor sequences learning although the progressive automatization of motor sequences involves successive offline consolidation periods (Doyon et al., 2003). Later consolidation stages of sequence learning in DCD seem to rely on atypical brain activation pattern with a lack of disengagement of regions generally involved in the early stages of learning (Biotteau et al., 2017). Future studies should investigate these later stages of procedural learning in DCD and complete behavioral data with neuroimaging data. Finally, to fully embrace the complexity of DCD, we did not exclude participants with associated neurodevelopmental disorders (such as DLD or SLD with reading or writing impairment). Studies objectively characterizing the severity of the symptoms related to different neurodevelopmental disorders such as SLD with impairment in reading or DLD might help to better characterize the profile of procedural learning performance related to each specific developmental disorder.

In conclusion, we observed reduced visuomotor sequences learning in children with DCD compared to TD peers after controlling for baseline RTs differences and intra-individual variability. Our results showed that children with DCD present reduced skills to learn sequential regularities in their environment although the discrimination and the learning of a repeating sequence remains possible (as highlighted by GPI and learning slope analyses respectively). This finding raises the importance of embracing the general functioning of the child while studying learning skills in neurodevelopmental disorders. This study also highlights associations between procedural learning skills and specific cognitive functions such as motor inhibition and verbal working memory, possibly relying on an atypical functioning of partly overlapping brain networks (Bo & Lee, 2013; Doyon et al., 2003; Janacsek et al., 2020).

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