Speech disfluencies in children with developmental dyslexia: how do they differ from typical development?

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Abstract

Background. Disfluency is a multifactorial concept that can be linked to several of the language production levels, both in typical and atypical populations. In children, the language system is still developing and few studies have explored disfluency patterns. In Typical Development (TD) in particular, studies have shown discrepancies according to the language being considered. In neurodevelopmental disorders, such as Developmental Dyslexia, it is still unclear whether the pattern of disfluency is similar to TD children.

Aims. Our study had two objectives. First, we analyzed the type of disfluencies and their evolution in French children aged 8 to 12 years old. Second, we compared the pattern of disfluency in DD and TD, and tested whether these difficulties were correlated with reading difficulties.

Methods & Procedures. 25 children with DD and 21 children with TD aged from 8 to 12.6 years were compared based on an autobiographical oral narrative. Seven types of disfluencies were coded: part-word repetitions; repetitions of monosyllabic words; other types of repetitions (words and phrases); filled pauses; revisions-substitutions; revisions-additions; abandoned utterances. We compared the proportion of each disfluency in DD and TD. Spearman correlations were then performed between disfluencies, reading performances and age.

Outcomes and Results. Our results showed that both DD and TD children mainly produced filled pauses, repetitions of monosyllabic words, and substitutions. In both groups, children had a high rate of disfluency (>10%). Correlations with reading performance were significant in the TD group only.

Conclusions & Implications. Our study showed that DD in not characterized by a specific pattern of disfluency, and the type of disfluencies produced was stable in children aged from 8 to 12 years old. In contrast to other languages, our study suggest that French-speaking children have a high rate of disfluency. In other words, disfluency should be interpreted with caution in DD, given that TD children also have a high rate of disfluency. It seems important to adapt the pathological threshold of disfluency to the language being spoken, in order to avoid an overestimation of the prevalence of these deficits in French-speaking children.

Keywords: disfluency, discourse, developmental dyslexia, language development

Introduction

The term ‘disfluency’ includes various phenomena such as filled or silent pauses, repeated words, and self-corrections. According to some estimates, from 2 to 26 disfluencies occur every 100 words in adults’ speech, depending on the phenomena that are analyzed as a disfluency (Fox Tree, 1995). Despite the high frequency of disfluencies, the question remains as to why speakers are so often disfluent. Within the language system, several of the language production levels may be involved in the production of disfluencies. It has been shown, for example, that hampering the conceptual generation of a message (Schnadt & Corley, 2006) or the initial stage of lexical access (Pistono & Hartsuiker, 2021) affected the rate of disfluencies. However, less is understood about children’s speech, children being still developing their language skills. Indeed, so far, only limited data are available about the frequency and types of disfluencies, and the developmental path of speech disfluencies in typically developing children. Because of this lack of empirical study, it is also difficult to analyze disfluencies related to neurodevelopmental disorders that affect language skills, such as developmental dyslexia.

In typical development (TD), most studies compared the proportion of typical disfluencies and stuttering-like disfluencies (SLD) in children’s speech, in order to identify early stuttering and predict persistent stuttering (suggesting a criterion of 3% SLD to discriminate stuttering). Stuttering-like disfluencies are usually accompanied by physical tension and last longer than typical disfluencies (Lickley, 2017). They include part-word repetitions, monosyllabic word repetitions, blocks and broken words, while typical disfluencies include phenomena like filled pauses, multisyllabic word repetitions, phrase repetitions, revisions and abandoned utterances (Jansson-Verkasalo et al., 2021). Jansson-Verkasalo and colleagues analyzed SLD and typical disfluencies in typically developing Finnish-speaking children aged 2 to 4 and 6 to 9 years old. They did not find any significant difference in the total frequency of disfluencies in children ages 2 to 4. In older children, they found a peak of disfluency in 6 years old compared to older groups. In each group, few SLD were produced in comparison to typical disfluencies but 57% of children scored above the criterion of 3% SLD per 100 words. It therefore seems that this threshold does not hold for all languages. Similarly, Leclercq et al., (2018), investigated disfluency in French-speaking children aged 4. They included SLD (i.e., part-word/sound-syllable repetitions, sound prolongations, blocks, broken words and monosyllabic word repetitions) and typical disfluencies (i.e., multisyllabic word repetitions,

phrase repetitions, revisions and filled pauses). The most common types of disfluencies were filled pauses, followed by revisions, monosyllabic word repetitions and phrase repetitions. The least frequent types were multisyllabic word repetitions and part-word repetitions, followed by broken word, sound prolongations and blocks. The total amount of disfluency was quite substantial (10% on average) in this group. Additionally, when including all monosyllabic word repetitions in the SLD, 40% of the children produced more than 3% SLD. This proportion drops when only considering monosyllabic word repetitions with 3 or more iterations as a SLD. It therefore seems that French-speaking children have a high rate of both typical and SLD. Finally, Carlo & Watson, (2003), compared disfluency production in Spanish-speaking children aged 3.5 to 4 and to 5 to 5.5 years old. When analyzing the total frequency of disfluencies, they revealed no main effects nor interaction for age or gender. Revisions, interjections (i.e., filled pauses), and monosyllabic word repetitions were the most frequently observed speech disfluencies, which is quite similar to what Leclercq et al. found in French.

In other words, it seems that the frequency of disfluency is more influenced by the language than the age group in TD. Additionally, besides stuttering, few studies investigated the effect of neurodevelopmental disorders on disfluency production. Balčiuniene & Kornev (2016), showed that children with specific language impairment produced less filled pauses but more part-word repetitions than typically developing children. To our knowledge, no study looked at the effect of developmental dyslexia on disfluency production. Developmental dyslexia (DD), is characterized by disorders affecting reading acquisition (i.e., deficit in precision and/or fluency in written word recognition, which is not explained by a neurological, sensory or intellectual dysfunction (ICD-10, World Health Organisation, 1992 and DSM-5 American Psychology Association, 2013). As a consequence, several “secondary” consequences arise, such as problems in reading comprehension and reduced reading experience, which can impact lexical development and background knowledge (Cappelli et al., 2022). Few studies investigated language production in DD. They showed that DD children (Hanly & Vandenberg, 2010) and adolescents (Faust & Sharfstein-Friedman, 2003) experience more tip-of-the tongue states during naming tasks, which parallels their problems at the level of phonological representation. Indeed, tip-of-the tongue states refer to situations during which one is unable to retrieve a word while being certain to know it. They occur when semantic and lexical representations corresponding to a word are activated, causing a strong feeling of knowing the word, but activation of phonological information about the word is incomplete, resulting in partial phonological activation (see Burke et al., 2018 for a review). These phenomena are

largely studied in healthy aging. According to some authors, disfluencies during discourse production are a manifestation of these word retrieval difficulties (e.g., Kavé & Goral, 2017). Besides, in a small sample of 9 to 10 years old dyslexic children, limitation in abilities to produce complete narratives in a picture-based narrative task was put forward (Kornev & Balčiūnienė, 2015). The capacity to properly tell a story was also partly related with executive functioning in children dyslexia (Fisher et al., 2019). We suppose that the difficulty to elicit elements within narrative discourse whether related to lexical access limitations, lack of exposure to written stories or working memory deficit could be associated with more disfluencies. We can therefore predict than children with DD will exhibit a high rate of disfluencies. However, whether the proportion of disfluencies or the type of disfluencies produced differ from TD remains to be known.

The current study will compare DD and TD aged 8 to 12 years old on disfluencies produced during an autobiographical narrative. We will first analyze disfluency patterns, to examine whether filled pauses, monosyllabic word repetitions, and revisions are also the most common type of disfluency in older children (i.e., as suggested by Leclerq et al., 2018). Then, we will test whether DD children produce more of these disfluencies, or other types of disfluencies. Finally, we will test whether disfluencies correlate with difficulties that are typical of DD, namely reading fluency and precision.

Methods

Participants

25 Children with DD and 21 children with TD participated in the study. They were aged 8 to 12 years old and 6 months, right-handed and native French speakers (DD group mean age = 120 months, 12 girls and 13 boys; TD group mean age = 121 months, 10 girls and 11 boys). Children with DD were referred by the Regional Centre for Learning Disabilities Diagnosis (from [redacted]) or by external therapists (e.g., speech therapists). They had normal or corrected-to-normal eyesight, normal hearing and they had no history of birth complication, neurological or psychiatric disorder (epilepsy or Autism Spectrum Disorder) and no psychotropic drug treatment.

Children were recruited based on the following criteria:

- DD group: performance regarding the speed or accuracy of reading isolated words was less than or equal to a standard deviation of −1.5 on the ODEDYS test (Jacquier-Roux et al., 2005) and speed or accuracy of reading performance (text reading) was less than or equal to a standard deviation of −1 (Alouette-R test, Lefavrais, 2005)
- TD group: performance in the reading test (cutoff Alouette-R test and ODEDYS test (speed or accuracy): ≥ −0.5 SD).

All children had typical performance in oral language (Linking Words subtest of EVAC test, Flessas & Lussier, 2003 and Ecosse test; Lecocq, 1996 > −2 SD), in motor abilities (cutoff M-ABC: ≥15th percentile), a normal intellectual functioning (cutoff Similarities and Picture Concepts subtests, French-language version of the WISC-IV, Wechsler, 2005: scaled score ≥7), and no associated ADHD (ADHD questionnaire, diagnostic criteria DSM-5, American Psychology Association [APA], 2013: cutoff as « hyperactivity/impulsiveness » as « inattention »: <6/9).

The study was approved by an institutional ethics committee and conducted in accordance with the Declaration of Helsinki. This study obtained ID-RCB registration [redacted] We obtained informed oral and written consent from the parents and their children.

Material

Task

To elicit connected-speech production, children were asked to recall a holiday memory. They were given the following instruction: "*Tell me about a memory of a holiday with as many details as possible. I was not there with you, and I would like to imagine it as if I were there*". The accuracy of the memories was checked with the parents to ensure the authenticity of the statements.

Discourse analyses

Participants' oral productions were recorded and manually and orthographically transcribed. Each transcript was checked by another rater.

Seven types of disfluencies were coded (Table 1): part-word repetitions; repetitions of monosyllabic words; other types of repetitions (words and phrases); filled pauses; revisions-substitutions; revisions-additions; abandoned utterances. Disfluencies were normalized on the number of words produced per participant. Other stuttering-like disfluencies than part-word repetitions and repetitions of monosyllabic words were not included because of their low occurrence.

Table 1. Example of each kind of analyzed disfluency.

Disfluency	**Example**
Part-word repetitions	"*we went to the* ***bea- beach***"
Repetitions of monosyllabic words	"*we went* ***to to*** *the beach*"
Other repetitions	"*we went* ***to the to the*** *beach*"; "*I* ***forgot forgot*** *my jacket*"
Filled pauses	"*we* ***uh*** *went to the beach*"
Substitutions	"*I* ***fall fell*** *from the chair*"; "*there were* ***shrimp- no lobsters***"
Additions	"*I was with* ***my sister my younger siste****r*"
Abandoned utterances	"*He talked about // but I was hungry*"

Analyses

Disfluency variables in DD and TD were compared using one tail t-tests.

To test whether disfluencies correlate with difficulties that are typical of DD, Spearman correlations were performed between variables that differed between the two groups and reading performance (accuracy and time, as measured with the Alouette-R test). To test whether the rate of disfluency varied between participants aged 8 to 12 years old, Spearman Correlations were performed between each type of disfluency and participants' age (in months).

Results

Descriptive

In both groups, filled pauses were the most common type of disfluency, followed by substitutions and repetitions of monosyllabic words (Table 2, in descending order). Additionally, Chi2 analyses showed that the proportion of each disfluency was comparable across groups (X^2 (6, N = 813) = 2.7, p = .07, Figure 1).

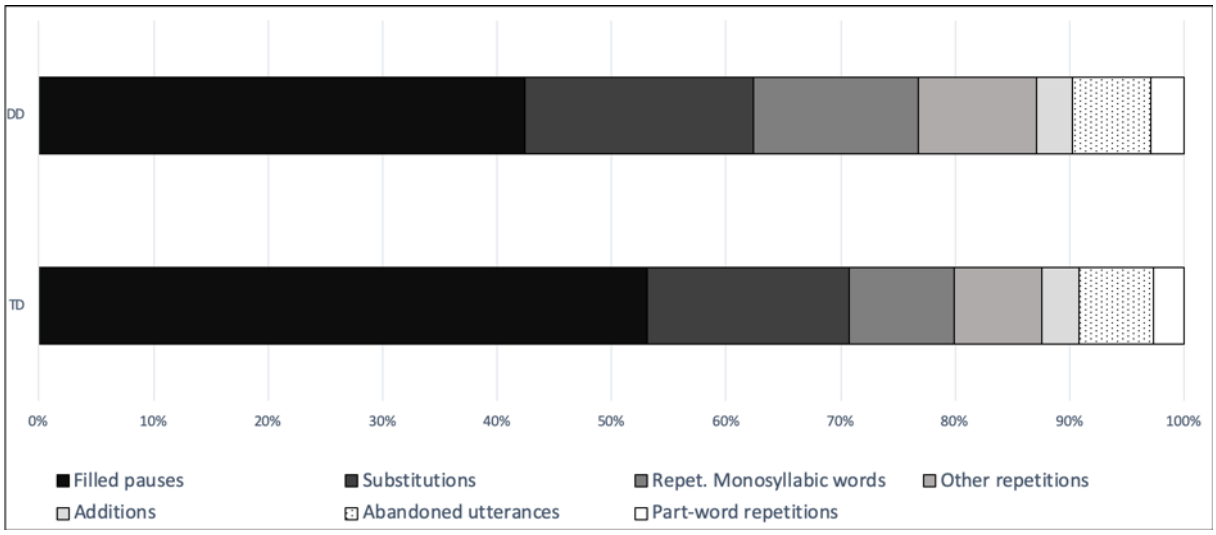

Figure 1. Proportion of each disfluency in DD group (upper band) and TD group (lower band).

Intergroup comparisons

The length of the narratives, measured in number of words, did not differ between groups (TD group: 191.5±104; DD group: 156.4±77; p=0.2). DD participants produced more substitutions (t(44)=-1.74; p<0.05) and more repetitions of monosyllabic words (t(44)=-1.73; p<0.05) than TD. Other variables did not significantly differ between groups (see Appendix).

Table 2. Descriptive results of disfluency production in each group (Mean ± SD).

	TD	**DD**	**p value**
Filled pauses	6.324 ± 4.208	5.274 ± 3.441	ns
Substitutions	1.422 ± 1.029	2.269 ± 2.038	<0.05
Repetitions of monosyllabic words	0.924 ± 1.038	1.624 ± 1.577	<0.05
Other repetitions	0.670 ± 0.755	0.929 ± 1.141	ns

Additions	0.327 ± 0.611	0.272 ± 0.404	ns
Abandoned utterances	0.537 ± 0.709	0.791 ± 0.775	ns
Part-word repetitions	0.255 ± 0.381	0.251 ± 0.523	ns
All disfluencies	10.54 ± 4.7	11.51 ± 4.5	

Correlations

Repetition of monosyllabic words and substitutions were not correlated with reading performance in the DD group. In the TD group, reading time was negatively correlated with repetitions of monosyllabic words, meaning that children who were slower readers produced more of these repetitions during their narrative. Age was not correlated with disfluency production, in any group (see Appendix).

Table 3. Correlations between disfluency, reading and age in each group. Significant results are in bold.

Variable		Reading accuracy	Reading time
DD Group			
	p-value		
Reading accuracy	Spearman's rho	—	
	p-value	—	
Reading time	Spearman's rho	**0.593**	—
	p-value	**0.002**	—
Repetitions monosyllabic	Spearman's rho	-0.161	-0.058
	p-value	0.443	0.784
Substitutions	Spearman's rho	0.210	0.254
	p-value	0.313	0.220
TD Group			
	p-value		
Reading accuracy	Spearman's rho	—	
	p-value	—	
Reading time	Spearman's rho	**0.736**	—

	p-value	**< .001**	—
Repetitions monosyllabic	Spearman's rho	-0.301	**-0.527**
	p-value	0.185	**0.014**
Substitutions	Spearman's rho	-0.009	-0.211
	p-value	0.971	0.358

Discussion

The current study aimed at analyzing the type and frequency of disfluency produced by TD and DD children aged 8 to 12 years old. It showed that both groups mainly produced filled pauses, repetitions of monosyllabic words, and substitutions. The DD group did not produce a different pattern of disfluency than the TD group. Additionally, disfluency production was correlated with reading performance in TD only.

In line with the literature based on French-speaking children we found that filled pauses, repetitions of monosyllabic words, and substitutions were the most frequent type of disfluency. Similar to Leclercq et al. (2018), we also found a substantial amount of disfluency in both groups (>10%), which suggest that French-speaking children have a high rate of disfluency. The current study, however, was conducted among children who were older than previous studies (French-speaking children aged 4: Leclercq et al., 2018; Spanish-speaking children aged 3 to 5: Carlos and Watson 2003; Finnish-speaking children aged 2 to 9: Jansson-Verkasalo et al., 2021). This suggests that disfluency patterns are quite stable across age groups, which was reinforced by a lack of correlation between disfluency and age in our study. Contrary to previous studies, we also distinguished revisions that were substitutions from the ones that were additions. By doing so, we showed that substitutions are highly frequent compared to additions. These two types of revisions do not have the same function. Indeed, additions reflect discourse-level revisions, aiming at bringing a precision or solving an ambiguity (e.g., “the box…the big one”) while substitutions of words are more related to lexical-level revisions (e.g., “the horse…the pony”). Current results therefore indicate that DD and TD children aged 8 to 12 produce few discourse-level revisions.

Most importantly, DD children produced the same types of disfluency as TD children. They produced slightly more repetitions of monosyllabic words and more substitutions than TD but did not differ on the type of disfluency produced. This shows that their phonological difficulties did not lead to other disfluencies. In particular, it did not lead to SLD, that did not occur in our corpus (e.g., none of them produced blocks such as "m#...mother" or prolongations such as "ssssunday"). These results argue against the covert repair hypothesis in the context of DD. The covert repair hypothesis (CRH, Postma & Kolk, 1993) has been developed to explain both typical and stuttering-like disfluencies. It assumes that problems in the timing of phonological encoding result in many errors in a representation of internal speech (in the selection of correct phonemes). A self-monitoring system will often detect these errors before they are articulated but these covert repairs will disrupt the fluent delivery of speech, leading to different types of disfluencies. Because of problems arising at the level of phonological representations in DD, the CRH would predict more SLD in this group (because of a higher proportion of errors related to the selection of incorrect phonemes). On the contrary, results suggest that common repair mechanisms are at stake in DD and TD, leading to the production of filled pauses, repetitions of monosyllabic words, and substitutions. Nonetheless, repetitions of monosyllabic words and substitutions are informative about DD, as they were significantly more frequent in this group. As mentioned earlier, substitutions usually reflect lexically-driven revisions. They increase with lexical access difficulties (Pistono & Hartsuiker, 2021) or in populations experiencing word-finding difficulties, such as healthy aging (Schmitter-Edgecombe et al., 2000) or Alzheimer's disease (Pistono et al., 2021). Given that some studies indicated that DD leads to an increase of tip-of-the-tongue states (Faust & Sharfstein-Friedman, 2003), substitutions may reflect such difficulties in discourse production. According to many authors, repetitions are also a proof of word-finding behavior (Schmitter-Edgecombe et al., 2000), in particular when repeated words are grammatical words, which are usually monosyllabic words (Morel & Danon-Boileau, 1998). Further work is however required to analyze these phenomena together with other measures of word-finding difficulties in DD, in order to understand the underlying cause of these disfluencies. In particular, previous work (Faust & Sharfstein-Friedman, 2003) showed correlations between tip-of-the-tongue-states and reading skills, indicating that DD participants (aged 12 to 14.6 years old) who achieved higher scores on reading tests also gave more correct responses and less tip-of-the-tongue states on a confrontation naming task. On the contrary, the current study found correlations between reading performance and disfluency measures in the control group only. This suggests that in younger children with DD, disfluencies do not only reflect difficulties in accessing a word's phonological code (as in a tip-

of-the-tongue state) but may also reflect other difficulties, such as discourse planning difficulties.

Limitations and future directions

This lack of correlations between reading skills and disfluency production reflects the multifactorial nature of disfluencies. In particular, previous work has shown that the underlying function of disfluency differ depending on the type of task: in Alzheimer's disease the production of pauses was correlated with patients' lexical-semantic performance in a picture-based narrative but these phenomena were correlated with their anterograde memory performance in a personal narrative (Pistono et al., 2019). It therefore seems that the task used to assess oral language skills plays a role. Here, we used autobiographical narratives (holiday memories), which may cause more disfluencies than a conversation or picture-based narrative due to a higher planning demand (as shown in Pistono et al., 2019 in Alzheimer's disease). Even though personal narratives have a high ecological validity and are less stressful for participants, other tasks may be more relevant to strictly assess language processing (i.e., storytelling or picture description). Additionally, other variables could be considered, targeting phonological encoding more specifically. It may be informative, for example, to analyze the type of words that are produced in TD and DD, such as their lexical frequency, length, phonological complexity, etc. Moreover, the current study treated age as a continuous variable in each group (contrary to other studies who compared close age groups), which could reinforce the lack of differences due to age. However, future work should conduct similar analyses, using a larger age span to get a better understanding of how each disfluency evolves across age.

Clinical implications

The current study showed interesting results for speech therapists, to guide patient care. First, our results are in line with Leclercq et al., 2018 and reinforce the idea that disfluency rate varies with language-typology, since we also found a high rate of disfluency in French-speaking children. Moreover, the presence of disfluencies in the narratives of preadolescents should not be considered as language dysfunction. Indeed, we observed the same rate of disfluencies between 8 and 12 years old as Leclercq and colleagues found in children aged 4. Lastly, the pattern of disfluency what quite similar in TD and DD (i.e., the same types of disfluency were produced), which is calling for cautiousness when interpreting disfluency in children with DD.

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