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Communicating care

La communication à coeur

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The Co-Occurrence of Possible Developmental Coordination Disorder and Suspected Childhood Apraxia of Speech

La cooccurrence d'un potentiel trouble développemental de la coordination chez les enfants soupçonnés d'avoir une dyspraxie verbale

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Abstract

Childhood Apraxia of Speech is a communication disorder characterized by deficits in planning and programming speech motor movements. Developmental Coordination Disorder is a neurodevelopmental disorder affecting the ability to plan and execute motor movements. The co-occurrence of Developmental Coordination Disorder and Childhood Apraxia of Speech is unknown. This study explored whether the prevalence of possible Developmental Coordination Disorder in a population of children with suspected Childhood Apraxia of Speech would be greater than the occurrence of Developmental Coordination Disorder in the general population. A sample of 35 children with suspected Childhood Apraxia of Speech was recruited and parents completed a Developmental Coordination Disorder screening questionnaire. Results indicated that children with suspected Childhood Apraxia of Speech were identified as being at greater risk for also having possible Developmental Coordination Disorder than are children in the general population for Developmental Coordination Disorder. The percentage of children (49%) with co-occurring suspected Childhood Apraxia of Speech and possible Developmental Coordination Disorder in the sample was significantly greater than the percentage of children (9%) with Developmental Coordination Disorder in the general population. Results support the need for a population-based prevalence study. Outcomes support the need for speech-language pathologists to implement multidisciplinary practice, early identification, and early intervention for this population.

Abrégé

La dyspraxie verbale est un trouble de la communication caractérisé par des déficits dans la planification et la programmation motrice des mouvements de la parole. Le trouble développemental de la coordination est, quant à lui, un trouble neurodéveloppemental qui affecte la planification et l'exécution motrice des mouvements. Or, la présence (ou absence) d'une cooccurrence de ces troubles chez un même individu n'est pas connue. La présente étude a exploré si la prévalence d'un potentiel trouble développemental de la coordination était plus élevée chez une population d'enfants soupçonnés d'avoir une dyspraxie verbale que chez des enfants provenant de la population générale. Trente-cinq enfants soupçonnés d'avoir une dyspraxie verbale ont été recrutés et il a été demandé à leurs parents de remplir un questionnaire de dépistage du trouble développemental de la coordination. Les résultats ont montré que les enfants soupçonnés d'avoir une dyspraxie verbale étaient plus à risque que les enfants provenant de la population générale de se faire également identifier un possible trouble développemental de la coordination. Le pourcentage d'enfants de l'échantillon chez qui était noté un possible trouble développemental de la coordination (49%) était significativement plus élevé que celui des enfants provenant de la population générale (9%). Ces résultats supportent la nécessité d'effectuer une étude de prévalence portant sur l'ensemble de la population. Ces résultats supportent également le rôle des orthophonistes dans la mise en œuvre d'une pratique multidisciplinaire, ainsi que dans le dépistage et l'intervention précoce du trouble développemental de la coordination chez les enfants soupçonnés d'avoir une dyspraxie verbale.

Childhood Apraxia of Speech

Childhood Apraxia of Speech (CAS) is a communication disorder characterized by deficits in planning and programming speech motor movements. CAS has been defined as

> A neurological childhood speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits.... The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody (American Speech-Language-Hearing Association [ASHA], 2007, para. 3).

The prevalence of CAS is currently unknown (ASHA, 2007). Clinical referral data suggests that CAS occurs in approximately 1–2 children per 1,000 (Shriberg, Aram, & Kwiatkowski, 1997) and in about 3%–4% of children with speech sound disorders (Delaney & Kent, 2004). Children with CAS can present at many different ages and in many different ways but are inclusively thought to have difficulty with planning and programming movements for speech (Ozanne, 2005). CAS appears to be the result of a neurological deficit, either idiopathic or as a result of impairment (ASHA, 2007).

The ability to describe CAS has improved, which has led to earlier identification. In practice, speech-language pathologists (S-LPs) often use clinical judgment and a combination of formal and informal measures to identify suspected Childhood Apraxia of Speech (sCAS; Strand, 2017). According to ASHA (2007), “at present, there is no validated list of diagnostic features of CAS that differentiates this symptom complex from other types of childhood speech sound disorders” (para. 3), but therapists often consider three consensus-based features: (a) inconsistent vowel and consonant errors, (b) lengthened and disrupted transitions between sounds and syllables, and (c) inappropriate prosody (intonation/stress).

In our clinical experience, S-LPs might not formally diagnose CAS, particularly in very young children, due to a lack of established diagnostic features. However, they may refer to a child as having sCAS when assessing speech and language skills, setting goals with families, and delivering treatment. Based on the literature, clinical judgments about sCAS are often made with the understanding that (a) there is a continuum of severity, (b) a child does not need to display every characteristic associated with CAS, and (c) characteristics of the disorder may change over time (Davis & Velleman, 2000). As part of diagnostic best practice, it is recommended that a period of treatment (i.e., 6 to 12 months) be delivered. If, after this period, speech intelligibility remains low and the motor control features of CAS remain, a firm diagnosis can be made (Davis & Velleman, 2000).

Developmental Coordination Disorder

Developmental Coordination Disorder (DCD) is a neurodevelopmental disorder in which “the acquisition and execution of coordinated motor skills is substantially below that expected given the individual’s chronological age and opportunity for skill learning and use” (American Psychiatric Association, 2013, p. 76). When motor impairments significantly impact a child’s ability to perform activities of daily living (i.e., self-care, academic productivity, and leisure activities at home, at school, and in the community) and cognitive disability, visual impairment, and other neurological conditions affecting movement are ruled out, a diagnosis of DCD can be considered. DCD occurs in 5%–6% of the population (American Psychiatric Association, 2013) with prevalence estimates varying by location from 5%–19% (Barnhart, Davenport, Epps, & Nordquist, 2003; Lingam, Hunt, Golding, Jongmans, & Emond, 2009; Slater, Hillier, & Civetta, 2010; Tsiotra et al., 2006; Zoia, Barnett, Wilson, & Hill, 2006) and 8% in Canada (Tsiotra et al., 2006). The cause of DCD is unknown, although neuroimaging studies have shown distinct brain differences in children with DCD compared to typically developing children (Zwicker, Missiuna, Harris, & Boyd, 2010, 2011, 2012).

In order to identify possible Developmental Coordination Disorder (pDCD), a screening tool may be used. Parent report has been established as a reliable source for helping to screen children not only for speech and language difficulties but also for motor difficulties—Webster, Majnemer, Platt, and Shevell (2005) used the Battelle Developmental Inventory and Gaines and Missiuna (2007) used the Child Development Inventory. The Developmental Coordination Disorder Questionnaire (DCDQ; Wilson et al., 2009) and the Little Developmental Coordination Disorder Questionnaire-Canadian Edition (Little DCDQ-CA; Rihtman, Wilson, & Parush, 2011; Wilson et al., 2015) are valid and reliable parent report tools for identification of pDCD in children 3 to 15 years of age.

The Relationship Between Childhood Apraxia of Speech and Developmental Coordination Disorder

Language disorders involve difficulty using and/or understanding words and sentences, while speech disorders involve difficulty articulating specific speech sounds. Studies show that children with developmental speech and language disorders often have motor skills that lag behind those of typically developing peers (see Rechetnikov & Maitra, 2009, for a review of speech-language disorders and motor skills) and that speech disorders—as compared to language disorders—may be more strongly correlated with poor gross motor skills (Visscher et al., 2010). More specifically, research has shown that both fine and gross motor impairments increase when speech or speech and language difficulties, as opposed to language only, are evident (Bishop, 2002; DiDonato-Brumbach & Goffman, 2014; Visscher et al., 2010; Visscher, Houwen, Scherder, Moolenaar, & Hartman, 2007). Visscher et al. (2010) concluded that a strong relationship exists between motor performance and speech as both require complex motor planning, programming, and execution.

The co-occurrence of motor difficulties and CAS has been explored further. In a longitudinal study of children at risk for familial CAS, Highman, Hennessey, Leitão, and Piek (2013) found that children at risk for CAS scored lower on measures of fine motor skills compared to children with no such family history. Hall (2000b) described possible associated factors with CAS reported in the literature, including gross-motor and fine-motor difficulties. Similarly, Davis and Velleman (2000) have described motor characteristics that may co-occur with CAS, including use of gestures to communicate, fine and gross motor delays, and motor clumsiness. Hodge (1998) described the parallels between DCD and CAS and questioned whether or not DCD could be initially detected in the speech motor system. In her theoretical model, Hodge (1998) discussed the possibility that DCD is the overarching sensorimotor disorder and is inclusive of a developmental speech coordination component. There has since been some evidence to suggest that the characteristics of DCD may also affect the speech motor system. For example, Ho and Wilmut (2010) found that children with DCD showed significantly different patterns of motor control, compared to typically developing children, during a complex sentence production task requiring a fast rate of speech.

A number of neuroimaging studies have indicated neurological differences between typically developing children and children diagnosed with DCD that could be contributing to motor planning and execution deficits. Biotteau et al. (2016) summarized these neuroimaging findings to conclude that the cerebellum, basal ganglia, parietal lobe, and limbic system could be involved. Brown-Lum and Zwicker (2017) summarized current functional magnetic resonance imaging studies that report (a) differences in brain activation, (b) differences in the corticospinal tract (motor) and the posterior thalamic radiations (sensory), (c) under-activation of the cerebellum, (d) deficits in the cerebellar network (i.e., connections to the frontal and parietal areas), and (e) differences in the cortical regions associated with working memory and executive function. Despite these neuroimaging studies, findings remain inconclusive in describing a unique structural or functional neural network signature for DCD. Regarding the neural correlates of speech production, Price (2012) reviewed research that highlighted the brain areas associated with heard speech, speech production, and reading, including the cerebellum's role in word generation. Broca's area and the supplementary motor area also have also been associated with speech movements, including planning and initiating sequential complex movements of speech articulators. Additionally, using functional magnetic resonance imaging, Redle et al. (2015) found that clients with persistent speech sound disorders demonstrated different patterns of brain activation than controls during a finger tapping task. Overall, the literature appears to suggest that both DCD and CAS may have similar underlying neural correlates, most notably the function of the cerebellum (Brown-Lum & Zwicker, 2017).

The treatment of both CAS and DCD may be more efficient and effective if conducted early and in a collaborative manner. In general, difficulties with communication and motor skills can significantly impact academic achievement and social participation; early assessment, diagnosis, and management by a multidisciplinary team is recommended in order to implement holistic, timely interventions and supports in an effort to minimize short- and long-term impacts (Hall 2000b, 2000c; Iverson & Braddock, 2011; Rechetnikov & Maitra, 2009; Visscher et al., 2010; Webster et al., 2005). More specifically, the research on CAS suggests that it is a complex motor speech disorder that may be best supported by a multidisciplinary team approach to treatment (Teverovsky, Bickel, & Feldman, 2009). As well, early identification and treatment of DCD is essential to the enhancement of participation in typical activities of childhood across all environments and the reduction of often devastating secondary consequences (i.e., anxiety, low self-esteem, poor self-efficacy, and limited participation; Engel-Yeger, 2015). Therefore, if a significant co-occurrence between these two disorders is found, then early identification and multidisciplinary treatment approaches targeting both disorders may prove to be both efficient and effective.

Present Study

To date, we are unaware of any study that specifically examines the co-occurrence of DCD and CAS. Because CAS is an impairment in planning and programming motor movements for speech, it follows that children identified as having sCAS may have broader underlying motor impairments and might therefore meet the criteria for DCD. It is possible that the prevalence of DCD in this population may be higher than in the typically developing population.

To address this gap in the literature, the purpose of this pilot study was to describe the co-occurrence of pDCD and sCAS in Alberta Health Services, Central Zone East, Children's Rehabilitation Services. We aimed to determine how many children between the ages of 3 and 15 years currently receiving services from community rehabilitation S-LPs in Alberta Health Services, Central Zone East, for sCAS would obtain scores on DCD parent questionnaires identifying them with pDCD. Central Zone East is a rural service area in Central Alberta where the general population is 122,057. Approximately 31,203 people in Central Zone East fell between the ages of 0–18 at the time of this study. The number of children aged 0–18 referred to Children's Rehabilitation Services for developmental concerns in 2016 was 1,423.

We hypothesized that the prevalence of pDCD in a population of children with sCAS would be greater than the occurrence of DCD in the general population. We also hypothesized that the proportion of children having both sCAS and pDCD in our present sample would be significantly greater than the proportion of children having the single diagnosis of DCD in the general population, implying that the sample population is unique.

The possible benefits from the outcomes of this project include (a) the advancement of our understanding about the possible co-occurrence of DCD and CAS, (b) the ability to characterize the potential involvement of a multidisciplinary team of clinicians in the early identification and treatment of sCAS and pDCD, and (c) to translate findings to daily practice locally, provincially, and nationally in a relatively short period of time.

Method

Participants

Ethics approval was received for this study from the Health-Research Ethics Board-Health Panel at the University of Alberta (Pro00067090). Potential participants were excluded from this study if they had been diagnosed with any known genetic, neurological, sensory, intellectual, or emotional disorder or deficit, cerebral palsy, a degenerative idiopathic motor disorder, or a traumatic brain injury, as per DCD differential diagnosis lists by Kirby, Sugden, and Purcell (2014) and Missiuna, Gaines, and Soucie (2006).

A convenience sample of children between the ages of 3 and 15 years receiving treatment for sCAS was recruited from Central Zone East. S-LPs working with these children identified them[1], asked their families if they would be interested in learning more about the study, and, if so, obtained informed consent to pass their name, contact information, and an sCAS checklist (developed for the purpose of this study and completed by the child's S-LP; see Appendix) to the research team.

Measures

The demographic form was created by the research team and included information about the targeted child and family related to age, sex, number of siblings, comorbidities (i.e., Attention Deficit Hyperactivity Disorder, autism, learning disability, language disability, dysarthria, executive function, joint hypermobility syndrome, anxiety, depression, overweight/obese, other), perceived physical activity level, household income, and level of parent education. Items on the demographic form were later used to describe the study population.

Physical activity level that appears in the demographics (see **Table 1** and **Table 2**) was parent-rated using the Canadian Physical Activity Guidelines (Canadian Society for Exercise Physiology, n.d.), which states that a child between the ages of 4 and 17 should participate in at least one hour of moderate to high intensity physical activity every day. Parents were asked to rate their perceptions of the targeted child's level of physical activity (not including gym class if school age). These perceptual categories included (a) very active (more than one hour of physical activity per day including extracurricular activities), (b) active (about an hour of vigorous physical activity per day), (c) somewhat active (2–4 hours of vigorous physical activity per week), and (d) prefers quiet activity (Canadian Society for Exercise Physiology, n.d.). Physical activity level was coded from the demographic form as 4 = *very active* (i.e., more than one hour of vigorous physical activity per day with involvement in extracurricular physical activities), 3 = *active* (i.e., about an hour of vigorous physical activity per day), 2 = *somewhat active* (i.e., 2–4 hours of vigorous physical activity per week), and 1 = *prefers quiet time* (e.g., reading, board games).

[1] Alberta Health Services Central Zone East speech-language pathologists (S-LPs) received six one-hour motor speech education sessions via Adobe Connect once a month from January to June 2016. Pre- and post-test results were compared and showed that S-LPs made significant gains in knowledge and confidence in providing all aspects (i.e., identification and intervention) of motor speech services. The results of this Motor Speech Education Project will be considered in establishing a motor speech education program for S-LPs in the province of Alberta (Forst, Meintzer, Klassen, & McAllister, n.d.).

Developmental Coordination Disorder questionnaires. Parents of 3–4 year-old children completed the Little Developmental Coordination Disorder Questionnaire-Canadian Edition (Little DCDQ-CA; Wilson et al., 2009), and parents of 5–15 year-old children completed the Developmental Coordination Disorder Questionnaire (DCDQ; Rihtman et al., 2011; Wilson et al., 2015). These questionnaires ask parents to use a 5-point Likert scale to compare their child's motor skills and coordination in everyday functional activities to those of their peers. Each questionnaire consists of 15 items that can be categorized into 3 factors (control during movement, fine motor, and general motor) for the DCDQ and 2 factors (fine motor and gross motor) for the Little DCDQ-CA (Wilson et al., 2009; Wilson et al., 2015). The Little DCDQ-CA and the DCDQ each yield a total score out of 75 and cutoff scores based on gender (Little DCDQ-CA) or age (DCDQ) indicate whether or not a child is suspect for DCD (Wilson et al., 2009; Wilson et al., 2015). The overall sensitivity for the DCDQ is 84.6% and specificity is 70.8% (Wilson et al., 2009). The Little DCDQ-CA reports sensitivity between 80% and 86% with specificity ranging from 49% to 63% (Wilson et al., 2015). The Little DCDQ-CA and the DCDQ were scored according to respective test protocols.

Suspected Childhood Apraxia of Speech checklist. The sCAS checklist was completed and submitted to the study team by the treating S-LP. Presently, there is no validated list of diagnostic features for CAS (ASHA, 2007). Therefore, the sCAS checklist developed and used in the present study included key characteristics based on the literature (see Appendix). This checklist was not developed as a tool to identify sCAS; rather, its purpose was to describe the speech characteristics of children already on caseload for sCAS.

A four-cluster arrangement of CAS descriptors was compiled based on Ozanne (2005) and guided by others (i.e., Apraxia Kids, n.d.; ASHA, 2007; Davis & Velleman, 2000; Fish, 2015; Hall, 2000a; Strand, 2017). *Cluster 1* characteristics describe phonological planning, *Cluster 2* characteristics describe motor programming, *Cluster 3* characteristics describe motor planning, and *Cluster 4* characteristics describe prosodic errors and negative history for babbling. Strand (2017) discussed several key characteristics of CAS (those "often present" and those "more discriminative") that help distinguish CAS from a more typical phonological impairment. Examples of these characteristics include a range of items from all four of the clusters in the compiled sCAS checklist, providing justification for their inclusion and the subsequent equal weighting given to each cluster in our analysis. An sCAS severity score was calculated by weighting equally all four clusters of the sCAS checklist to account for different numbers of items within each cluster. For each cluster, participants were assigned a percentage score based on the number of characteristics checked out of a given number of items in that cluster. Total scores from all four clusters were added and averaged for a total severity score percentage.

Procedure

Families who expressed interest in participating were contacted by a research team member who discussed the study, gained consent for participation, and sent a demographic form and age-relevant DCD questionnaire by mail or email. If the study team was unable to contact families on the first attempt or if families began but did not complete the study measures online, follow-up phone calls were made.

Data were collected from February to May of 2017. Following data collection, de-identified questionnaires were scored by the study team[2]. After surveys, demographic forms, and sCAS checklists were completed, the data were de-identified and assigned a participant number.

Data Analysis

Our main research question was to find out if children with sCAS would have a greater likelihood of being identified as also having pDCD compared to children being identified as having a single diagnosis of DCD in the general population. Our hypothesis was that the occurrence of pDCD in a population of children with sCAS would be significantly greater than the occurrence of DCD in the general population. To address this question, a binomial test was used to test the probability of pDCD in our present sample against DCD in the general population. In our study, two possible outcomes existed: having pDCD or not. We wanted to determine whether the likelihood of having pDCD was greater than chance alone. Based

[2] Families who participated were mailed a letter informing them whether their child's score fell in the probable Developmental Coordination Disorder range. If children scored in the probable Developmental Coordination Disorder range, parents were given the option to discuss further occupational therapy and physical therapy follow-up and assessment if interested.

on the literature, the prevalence of DCD in the general population is 5%–6% (American Psychiatric Association, 2013); we used the more conservative population 9% prevalence value for this test (Slater et al., 2010), as choosing this higher estimate allows for a more cautious comparison of our current study sample to the population sample.

Our second hypothesis was that our sample of children with sCAS and pDCD would reflect a different population group than individuals having only DCD identified in the general population. We employed a two-sample *z* test to compare the sample proportion of children with pDCD to the proportion of DCD in the general population.

Results

Participants

Parents of 35 out of 63 identified children participated, resulting in a response rate of 61.4%. Of the participating children, 31 were boys and 4 were girls. Demographic data relevant to these participants are detailed in **Table 1** and **Table 2**. The pDCD scores (representing total score on the Little DCDQ-CA or DCDQ) and the sCAS scores (including total sCAS score from the entire checklist and respective cluster scores, both as a percentage) are also listed.

Probability Statistics

In the present study, 17 of 35 participants scored below the cutoff for pDCD. We found that the probability of having exactly or more than 17 cases of pDCD in a sample of 35 is significantly greater than the probability of having a single diagnosis of DCD in the general population ($p < .0001$, 1-tailed). Thus, we can accept our hypothesis stating that the occurrence of pDCD in a population of children with sCAS is greater than the occurrence of a single diagnosis of DCD found in the general population.

Next, we wanted to determine whether or not 17 of 35 cases were significantly different from 10,985 of 122,057 (9% of the Central Zone East population). A *z* score of 8.3, $p < .0001$, indicated that the percentage of children (49%) that have co-occurring sCAS and pDCD in the present sample is significantly different than the percentage of children (9%) in the general population (see **Figure 1**). The 95% confidence intervals depicted in **Figure 1** suggest that the sample of children with sCAS and pDCD does not overlap with the general population of DCD and, therefore, likely describes a distinct group. Thus, we can accept our hypothesis that the two populations (i.e., the study population and general population) are distinctly different.

Table 1

Demographic Information for the Younger Group (3–4 Year Olds; *n* = 18): Little DCDQ-CA

	Age (months)	DCD Score	sCAS Total Score (%)	Cluster 1 Score (%)	Cluster 2 Score (%)	Cluster 3 Score (%)	Cluster 4 Score (%)	Physical Activity (Range = 1 to 4)	Number of Comorbidities
M	47.44	64/75	40.47	69.75	31.11	54.70	6.35	3.39	0.72
SD	7.22	10.98	17.11	25.79	27.63	24.02	13.17	0.78	0.57
Range	36–59	37–75	14.32– 77.14	11.11–100	0–80	30.77–100	0–28.57	2–4	0–2

Note. Cutoff score for "Suspect DCD" is 68 or below. A total of 56% of younger children fell below the cutoff score. DCD = Developmental Coordination Disorder; Little DCDQ-CA = Little Developmental Coordination Disorder Questionnaire-Canadian Edition; sCAS = suspected Childhood Apraxia of Speech.

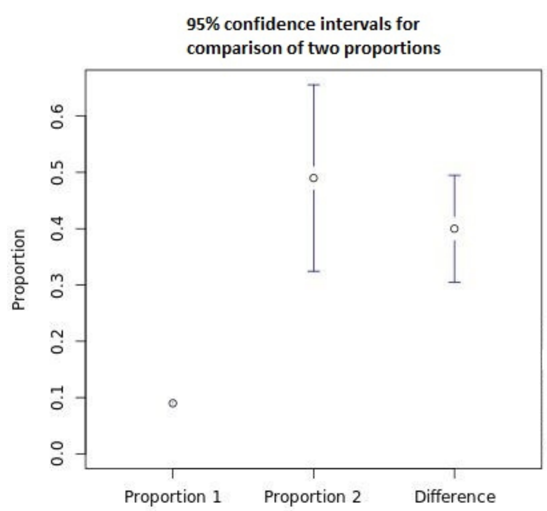
Table 2

Demographic Information for the Older Group (5–15 Year Olds; *n* =17): DCDQ

	Age (months)	DCD Score	sCAS Total Score (%)	Cluster 1 Score (%)	Cluster 2 Score (%)	Cluster 3 Score (%)	Cluster 4 Score (%)	Physical Activity (Range = 1 to 4)	Comorbidities
M	89.17	54.2/75	42.80	58.17	40	52.03	21.01	3.24	0.59
SD	25.79	12.33	19.18	29.80	27.39	28.85	24.28	0.83	0.62
Range	61–159	32–73	7.48–85.93	0–100	0–80	7.69–100	0–71.43	1–4	0–2

Note. Cutoff score for "Suspect DCD" is 46 or below for 5–7.11 years, 55 or below 8–9.11 years, 57 or below for 10–15 years. A total of 41% of older children fell below the cutoff score. DCD = Developmental Coordination Disorder; DCDQ = Developmental Coordination Disorder Questionnaire; sCAS = suspected Childhood Apraxia of Speech.

Figure 1



Two sample *z* test for the comparison of two proportions. Proportion 1 represents the likely proportion of Developmental Coordination Disorder cases in Central Zone East based on general population using a conservative estimate of 9% (range 5%–9%; Slater et al., 2010). Proportion 2 represents the proportion of possible Developmental Coordination Disorder cases in the study sample of children with suspected Childhood Apraxia of Speech. The "Difference" represents the difference between the two proportions. The 95% confidence intervals are represented by the blue error bars.

Discussion

Identification of Possible Developmental Coordination Disorder in Suspected Childhood Apraxia of Speech

This pilot study provided an initial evaluation of the co-occurrence of sCAS and pDCD in children ranging from 3–15 years of age. Data from the present study sample of children with sCAS showed a significantly higher proportion of pDCD (49%) than the 5%–9% (Slater et al., 2010) of individuals with DCD in the general population. Given the proportion confidence intervals for our sample and for the general population, it is unlikely that we are simply identifying individuals with DCD but rather a distinct sample of children with shared speech and coordination disorders.

These results support the need for a prevalence study on a larger population of children with sCAS in an effort to advance our understanding of the possible shared motor planning deficits between CAS and DCD, which will advance clinical best practice across the pediatric rehabilitation disciplines that specifically work with these children.

Implications

Most often, children who present with possible motor speech delays and disorders are first brought to the attention of S-LPs (Missiuna, Gaines, & Pollock, 2002). Based on the results of this study, it is important that S-LPs be aware of DCD, its prevalence in the general population, and its co-occurrence in children with motor speech disorders (i.e., sCAS or CAS). S-LPs may want to consider using screening tools such as the Little DCDQ-CA or

DCDQ as part of their initial screening protocol for sCAS. In light of the present results, a collaborative, multi-disciplinary (i.e., physical therapy, occupational therapy, speech language pathology, therapy assistant) approach may be particularly important for early identification of DCD and for providing ongoing supports, treatment, and management of children identified as having sCAS and pDCD. Taking a holistic approach to client care is likely to result in gains in multiple motor domains.

Limitations and Future Directions

We have preliminary evidence that there is a co-occurrence between sCAS and pDCD. Because of the small sample size, it will be very important to apply this methodology to a larger population in order to increase statistical power and generalizability. In addition, it would be beneficial for S-LPs to know if there are any particular diagnostic features of sCAS that predict pDCD in order to make more effective clinical decisions regarding client care and to effectively determine when a multidisciplinary approach is needed. Having considered potential characteristics of sCAS and their co-occurrence with pDCD at the outset of the present study, we developed the sCAS checklist. The sCAS checklist used in the present study has not gone through rigorous psychometric development. Ideally, a validated sCAS checklist in combination with clinical impressions would trigger a referral for DCD screening as a best practice process. Further research is needed to validate this checklist as well as explore whether there are characteristics of sCAS that correlate with pDCD and/or predict scores on DCD parent questionnaires. Increased statistical power would also be needed to address these questions.

Another limitation of this pilot study is the use of the Little DCDQ-CA and DCDQ rather than a validated motor assessment along with diagnostic criteria to assess and diagnose DCD. The use of the DSM-5 diagnostic criteria (American Psychiatric Association, 2013), combined with a motor score determined by the application of a validated motor assessment could be used to further support the prevalence of DCD within the sCAS or CAS populations. Moreover, correlates of DCD and physical activity level (e.g., experience and environment) would be strengthened by using quantitative measures of physical activity (e.g., pedometer, fitness tracker) in combination with parent perceptions of activity levels.

Once statistical power has been achieved for this particular method and we have a better understanding of the shared motor deficits between DCD and CAS, translation of this process could take place in the form of training multidisciplinary teams in the context of identification, intervention, management, and support mechanisms, leading to best practice clinical guidelines for this population.

Conclusion

This pilot study was designed to explore the co-occurrence of pDCD in a sample of children with sCAS. Our findings revealed a significant probability that children identified as having sCAS may also have pDCD. These preliminary results have implications for early identification and multidisciplinary involvement to support these children.

This information has the potential to support S-LPs in the management of sCAS, support future recommendations for S-LPs that all children with sCAS should be screened for pDCD, and support a multidisciplinary approach to early identification, intervention to promote lifelong participation, and prevention of secondary consequences. S-LPs who do not work regularly with multidisciplinary team members might consider helping clients pursue other physical therapy and occupational therapy service options as needed. Results of this study may also support the need for tele-practice options when physical therapy and occupational therapy services are not immediately available for families.

The results of this preliminary study could be used to support the need for further research using formal assessment tools and/or validated checklists to examine the prevalence of pDCD in sCAS and even the prevalence of DCD in CAS. In studies with larger sample sizes, the specific sCAS criteria that account for variance in pDCD scores would merit investigation. S-LPs could use this information in screening clients and be diligent in looking for any red flags to necessitate screening for DCD. As a result of the high prevalence of pDCD in the present sample, possible genetic or neurological links between CAS and DCD may also be investigated.

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Disclosures

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Appendix

Survey Questions

Suspected Childhood Apraxia of Speech Checklist

Instructions: Please check off the indicators that you observed with each client that made you suspect they had Childhood Apraxia of Speech. You do NOT have to go back and rescreen any clients.

Cluster	*Description*	Check Box if Present or Observed
Cluster 1 Phonological Planning	Inconsistent production of same word	
	Sounds or words may disappear for a period of time during therapy	
	Correct production of a difficult word may occur but cannot be repeated	
	Speech may be “easy” one day and “hard” the next	
	Unusual, idiosyncratic error patterns (sometimes defying transcription!)	
	Increased errors with increased performance load (i.e., repetition of words, especially longer words; words in phrases or sentences)	
	Errors that cannot be explained in terms of common articulation or phonological process errors	
	Poor maintenance of phonotactic structure (permissible syllable structure, consonant clusters, and vowel sequences)	
	Vowel errors (limited repertoire of vowels; less differentiation between vowel productions; vowel errors, especially distortions)	
Cluster 2 Motor programming	Slow diadochokinetic rates	
	Poor sequencing ability of diadochokinetic tasks	
	May have difficulty with volitional nonspeech movements	
	Possible voicing errors	
	Possible resonance inconsistencies	
Cluster 3 Motor planning	Substitutions	
	Deletions	
	Additions	
	Distortions	

	Reversals of sounds in words
	Reversals of syllables in words
	Errors increase or change as number of repetitions increases
	Centralize vowels to “schwa”
	Spontaneous production of phonemes in words but unable to imitate
	Well-rehearsed “automatic” speech is easiest to produce; “on demand” speech is most difficult
	Use of phonemes in words that do not contain that phoneme, but errors on that phoneme in the appropriate context (e.g., “wasi” for “apple” but “bimpoe” for “window”)
	Some groping (e.g., trial and error movements on the imitation of single sounds) may be noted
	Prolonged pauses between phonemes, syllables, and words due to challenges with making smooth articulatory transitions from phoneme-to-phoneme or syllable-to-syllable; pauses and breaks between phonemes may give the child’s speech a staccato quality
Cluster 4 Prosodic differences No history of babbling	Rhythm and stress of speech are disrupted
	Apply stress to the wrong syllable of a word
	Use excessive equal stress by applying excessive stress equally to each syllable of a word, giving speech a robotic quality
	Use excessive equal stress on all or most words of a sentence, giving speech a monotone or staccato quality
	Apply stress to an inappropriate lexical item within a sentence
	Overall slow rate
	No history of babbling

Note. Checklist informed by Apraxia Kids (n.d.), American Speech-Language-Hearing Association (2007), Davis and Velleman (2000), Fish (2015), Hall (2000a), Ozanne (2005), and Strand (2017).

Analysis of Naming Errors in Healthy Aging, Mild Cognitive Impairment, and Alzheimer's Disease

Analyse des erreurs de dénomination dans le vieillissement normal, le trouble cognitif léger et la maladie d'Alzheimer

KEYWORDS
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Abstract

The aim of this study was to document the functional origin of anomia in mild cognitive impairment in comparison to Alzheimer's disease and healthy cognitive aging. An oral naming task of 260 pictures was administered to 20 individuals with mild cognitive impairment, 5 with mild Alzheimer's disease, and 15 healthy controls. The mean total number of errors and types of naming errors were compared across the groups. The effect of psycholinguistic parameters and the efficacy of semantic and phonological cueing were also analyzed. Results showed a significant difference among the three groups' total number of naming errors (Alzheimer's disease > mild cognitive impairment > healthy controls). Similar types of naming errors were found among the groups and mainly consisted of coordinate semantic paraphasias. Further, less familiar words were associated with greater error probability in all groups. Finally, based on error types, psycholinguistic parameters, and efficacy of cueing, the main origin of anomia was determined for each participant and different patterns were observed among the three groups. In healthy controls, the origin of anomia was lexical. In mild cognitive impairment, the origin of anomia was lexical for 60% and semantic for 40% of participants. In Alzheimer's disease, a degradation of fine and distinctive semantic features seems to be the main cause of anomia. Although the present data are limited due to small sample size, they will be useful in the development of appropriate interventions aiming to reduce anomia in the elderly.

Abrégé

L'objectif de la présente étude était de documenter l'origine fonctionnelle de l'anomie chez des individus ayant un trouble cognitif léger, lorsque comparés à des individus atteints de la maladie d'Alzheimer et des individus ayant un vieillissement cognitif normal. Pour ce faire, une tâche de dénomination orale, composée de 260 stimuli visuels, a été administrée à 20 individus ayant un trouble cognitif léger, 5 individus atteints d'une forme légère de la maladie d'Alzheimer et 15 individus ayant un vieillissement cognitif normal. Le nombre total d'erreurs de dénomination, ainsi que le type d'erreurs, ont été comparés. L'influence des propriétés psycholinguistiques des mots, ainsi que l'efficacité de l'indiçage phonologique et sémantique, sur la probabilité de commettre une erreur ont également été analysées. Les résultats ont révélé des différences significatives entre les trois groupes quant au nombre total d'erreurs de dénomination (maladie d'Alzheimer > trouble cognitif léger > vieillissement cognitif normal). Néanmoins, le type d'erreurs effectuées par les individus des trois groupes était similaire; il s'agissait principalement d'erreurs de type paraphasies sémantiques coordonnées. Ajoutons également que les mots moins familiers étaient associés à un plus grand risque d'erreurs dans les trois groupes. Enfin, en s'appuyant sur le type d'erreurs effectuées par les individus de chaque groupe, l'influence des paramètres psycholinguistiques et l'efficacité de l'indiçage, l'origine fonctionnelle de l'anomie a été déterminée pour chaque participant. Différents patrons ont été observés pour chacun des trois groupes. Chez les individus ayant un vieillissement cognitif normal, l'origine de l'anomie était principalement lexicale. Chez les individus ayant un trouble cognitif léger, l'origine de l'anomie était lexicale dans 60% des cas, alors qu'elle était sémantique dans l'autre 40% des cas. Chez les individus atteints de la maladie d'Alzheimer, la dégradation des caractéristiques sémantiques fines et distinctives semblait être la principale cause de l'anomie. Malgré les limitations dues à la taille de l'échantillon, les données recueillies dans le cadre de la présente étude seront utiles au développement d'interventions visant à réduire l'anomie des personnes âgées.

Alzheimer's disease (AD) is a neurodegenerative disorder leading to a progressive cognitive and functional decline. AD is typically preceded by a prodromal phase, the most frequent being mild cognitive impairment (MCI; Albert et al., 2011). Various cognitive symptoms can be found in MCI, but when it is associated with an underlying AD, a deficit in episodic memory is the core feature (Belleville, Sylvain-Roy, de Boysson, & Ménard, 2008; Hudon, Villeneuve, & Belleville, 2011). Impairments in other cognitive functions can also be observed, such as semantic memory (Belleville et al., 2008; Callahan et al., 2015; Joubert et al., 2008; Salmon, 2012), executive functions (Johns et al., 2012), visuospatial functions (Mitolo et al., 2013), and language (for reviews, see Taler & Phillips, 2008, and Tsantali, Economidis, & Tsolaki, 2013).

Compared to other cognitive functions, fewer studies have investigated language impairment in MCI. Yet, anomia (word-finding difficulties) is one of the most frequent complaints of people with MCI (Taler & Phillips, 2008). Anomia is usually assessed using a naming task where the participant has to produce the specific word corresponding to an object or image. According to the cognitive model proposed by Caramazza and Hillis (1990), oral naming of pictures involves the serial activation of several components including (a) pictographic analysis, (b) structural representations lexicon, (c) semantic system, (d) phonological output lexicon, (e) phonological buffer, and (f) articulatory system. The number of naming errors determines the presence of anomia, while the types of errors are essential to determine the functional origin of the impairment (Grima & Franklin, 2017).

In an oral picture naming task (e.g., "apple"), errors can be classified into several types (Bogliotti, 2012): (a) visual error, a perceptual error without a semantic link to the target word (i.e., "ball"); (b) superordinate categorical semantic paraphasia, naming the general category of the target word (i.e., "fruit"); (c) coordinate categorical semantic paraphasia, naming an object in the same category of the target word (i.e., "pear"); (d) associative semantic paraphasia, the production of a word with a semantic association to the target word, without regard to the grammatical class (i.e., "juice"); (e) visuosemantic error, an error that could be classified as perceptual but also as semantic (i.e., "peach"); (f) phonological paraphasia, a segmental error within the phonological form of the word (i.e., "papple"); (g) vague circumlocution, a summary description of the word that does not include any distinctive features of the object (i.e., "fruit with peel") and that is used as a compensatory strategy for anomia; (h) precise circumlocution, which is a more detailed description of the object (i.e., "fruit with peel, it could be red or green and it is used to make juice or pie") and that is also used as a compensatory strategy for anomia; (i) sequences of phonemic approximation, successive attempts to produce the target word (i.e., "an a, an ap, an apple"); and (j) neologism, the production of a word that does not belong to the lexicon of a specific language (i.e., "kivos"). It is also possible to have other types of manifestations like perseveration, the repetition of the same word in response to different target words (i.e., "bread" for several consecutive items); stereotypy, the repetition of a syllable, a word, or a fixed expression (i.e., "p, p, p" or fixed expression in the naming attempts); tip-of-the-tongue, the feeling that retrieval is imminent (i.e., "I know what it is, but I can't find the exact word"); and non-responses, that is when no answer is attempted (i.e., "I don't know").

According to the Caramazza and Hillis (1990) model, an impairment of pictographic analysis or structural representations lexicon could lead to the production of visual errors. An impairment at the semantic level (difficulty to access or retrieve the semantic feature of the target) could result in the production of non-responses, vague circumlocutions, or semantic paraphasias. An impairment at the lexical level (difficulty in the access or within the phonological output lexicon) could result in tip-of-the-tongue, production of precise circumlocutions, and phonological or semantic paraphasias (Chomel-Guillaume, Leloup, Bernard, Riva, & François-Guinaud, 2010). Finally, an impairment of the phonological buffer usually causes successive attempts to produce the target word through sequences of phonemic approximations.

In addition to the analysis of the error types, the analysis of the efficacy of semantic and phonological cues in a naming task increases the understanding of the functional origin of anomia (Nickels, 2001; Whitworth, Webster, & Howard, 2013). Indeed, when naming is facilitated by the production of the first sound of the target word (phonological cue), this leads to a lexical origin of anomia. In contrast, when naming is facilitated by the production of semantic features associated with the target word (semantic cue), the origin of anomia is more likely semantic. Psycholinguistic parameters, such as frequency, subjective frequency, and semantic category, can also have a notable influence on naming performance (Whitworth et al., 2013). Indeed, retrieval of the semantic features within semantic memory is influenced by subjective frequency (the frequency modulated by individuals' experience) and category (e.g., biological vs. manufactured), while retrieval of the phonological representations within the phonological output lexicon is influenced mainly by frequency (occurrence of a word, compared to other words in a specific language). Even if imageability (the ease with which a word evokes a visual or auditory picture) also influences the production of words, its impact is minimized in oral naming tasks because all visual stimuli typically imply a high level of imageability. Finally, word length has an impact in oral naming performance because the more sounds a word contains, the more difficult it is to maintain it in short-term memory (for more information see Chomel-Guillaume et al., 2010, and Whitworth et al., 2013).

According to the current body of research exploring anomia in MCI, biological items are more impaired than manufactured ones (Callahan et al., 2015; Duong, Whitehead, Hanratty, & Chertkow, 2006; Taler, Voronchikhina, Gorfine, & Lukasik, 2016), even if some authors found no impact of semantic category on performance (Laws, Adlington, Gale, Moreno-Martínez, & Sartori, 2007; Lockyer, Sheppard, & Taler, 2015). Quantitatively, anomia is less prominent in MCI than in AD (Balthazar, Cendes, & Damasceno, 2008; Lin et al., 2014). Results are less consistent when individuals with MCI are compared to healthy controls (HC). Indeed, some authors found that anomia was more prominent in MCI (Adlam, Bozeat, Arnold, Watson, & Hodges, 2006; Balthazar et al., 2008; Balthazar, Yasuda, Cendes, & Damasceno, 2010; Dudas, Clague, Thompson, Graham, & Hodges, 2005), while no difference between the two groups was found in other studies (Beinhoff, Hilbert, Bittner, Grön, & Riepe, 2005; Willers, Feldman, & Allegri, 2008).

Most studies analyzing the types of naming errors were conducted with AD participants (i.e., Barbarotto, Capitani, Jori, Laiacona, & Molinari, 1998; Cuetos, Gonzalez-Nosti, & Martínez, 2005; Gonnerman, Aronoff, Almor, Kempler, & Andersen, 2004; Lin et al., 2014; Silagi, Bertolucci, & Ortiz, 2015). Lin et al. (2014) concluded that a progressive degradation of the semantic system was responsible for anomia in AD, given the predominance of non-responses and semantic errors. However, they did not take into account the psycholinguistic parameters. There is no agreement on the origin of anomia in MCI. Some studies suggest a semantic origin (Adlam et al., 2006; Joubert et al., 2010; Willers et al., 2008), while others suggest a lexical-semantic one (Balthazar et al., 2008; Duong et al., 2006; Guidi, Paciaroni, Paolini, Scarpino, & Burn, 2015; Taler & Phillips, 2008), namely an impairment in both the semantic system and the phonological output lexicon. Until now, only two studies have documented the patterns of naming errors in MCI. Balthazar et al. (2008) compared the naming performances of individuals with MCI, AD, and HC. They found that the three groups were statistically different regarding number of correct answers, but had a similar pattern of errors: (a) coordinate semantic errors, (b) superordinate semantic errors, (c) circumlocutions, (d) visual errors, and (e) non-responses. The authors concluded that the observed errors came from a combination of partial degradation of the semantic system (predominance of semantic errors) and impairment in the phonological output lexicon and its access (effectiveness of phonological cueing). Willers et al. (2008) also compared the performance of MCI, AD, and HC participants. They found that the performance of participants with MCI was similar to those with HC regarding the number of correct answers, but better than participants with AD. Results also showed a similar pattern of error types in MCI and AD: (a) semantic errors, (b) non-responses, (c) visual errors, (d) other errors, and (e) phonological paraphasias. Based on these results, specifically the predominance of semantic errors and non-responses, the authors concluded that the naming errors were caused by an impairment of the semantic system.

Studies published to date have several limitations. First, there is a lack of precision in the classification of naming errors used by Balthazar et al. (2008) and Willers et al. (2008). Indeed, their classification did not take into account the difference between precise circumlocutions (precise expressions, including specific semantic features and generally associated with a lexical deficit) and vague circumlocutions (approximate or imprecise expression without specific semantic features, and generally associated with a semantic deficit; see Chomel-Guillaume et al., 2010; Whitworth et al., 2013). Moreover, psycholinguistic parameters (frequency, semantic category, subjective frequency) were not taken into account, despite the fact that they have an impact on oral naming (Whitworth et al., 2013) as described above. Finally, the authors used no objective measures to assess the integrity of visual recognition (e.g., object decision task), which limits the interpretation of the naming errors. Indeed, both studies used a picture-based oral naming task without determining that the participant had no visual agnosia. Also, the studies did not assess the integrity of the semantic system; however, an appropriate assessment could support the conclusion regarding the origin of anomia.

The aim of this study was to document the functional origin of anomia in participants with MCI in comparison to those with AD or HC. Specific objectives were to (a) quantitatively analyze the naming errors in the three groups to compare their performance to confirm the presence and severity of anomia and (b) qualitatively analyze the naming errors, taking into account the effects of psycholinguistic parameters and the effectiveness of semantic and phonological cueing. It was expected that the three groups would show different degrees of anomia following a continuum of severity (HC<MCI<AD). It was also expected that the analysis of naming errors combined with the effect of psycholinguistic parameters and cueing would highlight a lexical-semantic origin to explain the naming impairment in participants with MCI and AD (Balthazar et al., 2008; Duong et al., 2006; Guidi et al., 2015; Taler & Phillips, 2008).

Method

Participants

Initially, 60 participants were recruited: 12 with mild AD, 29 with MCI, and 19 HC. The study's final sample consisted of 40 participants aged 55 years and older: five with mild AD, 20 with amnestic MCI (14 single domain and six multiple domains), and 15 HC. Twenty participants were excluded from the initial sample because they had probable visuoperceptual difficulties according to the Object Decision subtest of the Birmingham Object Recognition Battery (Riddoch & Humphreys, 1993). Participants in the HC group were recruited from a database of cognitively and physically healthy older subjects and none had cognitive impairments or mental health disorders. Participants in the AD and MCI groups were referred by collaborating physicians or recruited through pamphlets distributed in medical clinics in Québec City (Canada). Participants with AD met the core clinical diagnostic criteria proposed by McKhann et al. (2011) and participants with MCI met the clinical diagnostic criteria of Albert et al. (2011). All participants in the MCI group had mild episodic memory impairment (with or without impairment in other cognitive domains).

For all participants, exclusion criteria were (a) neurodegenerative disorder (e.g., Parkinson's disease, multiple sclerosis), (b) history of moderate or severe traumatic brain injury or stroke, (c) interventions that may affect cognitive functioning (e.g., general anesthesia in the last 6 months, etc.), (d) delirium in the last 6 months or clinically significant psychiatric disorder according to the criteria of the DSM-IV-TR, (e) untreated or unstable medical or metabolic condition, (f) history of encephalitis or bacterial meningitis, (g) alcoholism or substance abuse in the last 12 months, (h) uncorrected visual or auditory problems, (i) incapacity to give consent to the study procedures, and (j) visual agnosia or probable visuoperceptual difficulties.

Material and Procedures

Two 2-hour sessions took place either at the research centre or at home, depending on the participant's choice. When sessions took place at the research centre, participants were offered a financial compensation of $20 per session for their travel expenses. The study procedures were approved by the Ethics Research Board of the Institut universitaire en santé mentale de Québec (approval #220).

Clinical and neuropsychological assessment. During the first assessment session, a clinical and neuropsychological battery was administered to confirm the diagnosis of AD or MCI in the clinical groups, or the absence of cognitive impairment in the HC group. The battery also aimed at verifying exclusion criteria. General cognitive functioning was assessed with the Montreal Cognitive Assessment (MoCA; Nasreddine et al., 2005; normative data by Larouche et al., 2015), depressive symptoms with the French version of the Geriatric Depression Scale (Bourque, Blanchard, & Vézina, 1990; Yesavage, 1988), and memory complaint with the Cognitive Complaints Questionnaire (*Questionnaire de plainte cognitive*; Thomas-Antérion, Ribas, Honoré-Masson, Million, & Laurent, 2004). Verbal episodic memory was evaluated with the 16-item Free/Cued Recall task (Van der Linden et al., 2004; normative data by Dion et al., 2015), and visual episodic memory with the immediate recall (3 minutes) of the Rey Complex Figure (Rey, 1960; normative data by Tremblay et al., 2015). The Rey Complex Figure was also used to assess visuoconstructive functions. Visual agnosia was evaluated using the Object Decision task (difficult test) of the Birmingham Object Recognition Battery (Riddoch & Humphreys, 1993). Semantic memory was assessed with the Pyramids and Palm Trees Test (Howard & Patterson, 1992; normative data by Callahan et al., 2010) and executive functions were assessed with the Stroop test of the D-KEFS battery (Delis, Kaplan, & Kramer, 2001). Finally, the battery included a task of phonemic (i.e., letters T, N, P) and semantic (i.e., animals) verbal fluency. For each group, results of the clinical and neuropsychological evaluations are presented in the Results.

Anomia assessment. During the second session, anomia was assessed for each participant using a computerized naming task (MS PowerPoint©) comprising the 260 visual stimuli of Snodgrass and Vanderwart (1980). The colour version of the pictures was used (Rossion & Pourtois, 2004). Frequency (n = 228 words), category (n = 260), and subjective frequency (n = 239) of the words were known for almost all stimuli (n = 260) of Snodgrass and Vanderwart. Those psycholinguistic parameters were found in the OMNILEX Canadian database from the University of Ottawa (Desrochers, 2006). Values for each parameter were the following: frequency (low, moderate, high), subjective frequency (low, moderate, high), and semantic category (biological, manufactured). Imageability and word length were not manipulated in this study because, as it was said previously, all visual stimuli typically imply a high level of imageability and word length's effect is related to a post-lexical treatment, at the phonological buffer level, a component that is not affected in AD.

For the naming task, participants were asked to name the objects that appeared in the centre of the computer screen. Responses were audio recorded. Two practice items, not included in the Snodgrass and Vanderwart (1980) images, were presented first to the participants. For each stimulus, participants had a maximum of 15 seconds to answer. If the answer was correct, the next stimulus was presented. If participants were unable to name an object after 15 seconds or gave a wrong answer, a semantic cue was given. For manufactured items (e.g., glass), the semantic cue always referred to the category and the use of the object (i.e., a kitchen tool used to drink), while for biological items (e.g., apple) the cue referred to the category and a perceptual characteristic of the object (i.e., a fruit that is generally red). Following the cue, participants had a maximum of 10 seconds to name the object. If the answer was correct, the next image was presented. However, if the answer was still wrong a phonological cue (first sound of the word) was given and participants had a maximum of 10 seconds to answer. If participants were still unable to give the correct answer, no feedback was provided and the next picture was presented.

Data Analysis

Error analysis. Incorrect spontaneous answers (i.e., before semantic or phonological cueing) were classified into 15 types of errors (Bogliotti, 2012). To ensure the error analysis was reliable, two independent raters analyzed types of errors made by the participants. To compare the sociodemographic, clinical, and neuropsychological characteristics of the three groups, analysis of variance (ANOVA) was used for continuous variables and Pearson's chi-square test was used for binary variables. An ANOVA was also used to compare the efficacy of semantic and phonological cueing between groups. An exact logistic regression model was used to evaluate the difference among the three groups' naming errors in spontaneous answers. An exact approach was used to take into account the small sample size of the AD group (Mehta & Patel, 1995). A mixed-effect Poisson model was then applied to compare the patterns of errors among groups. This model uses the logarithm on the number of errors as the link function, considers the participant as a random effect, and uses an offset corresponding to the logarithm of the total number of errors made by the participants. The sources of variance composing the fixed part of this model are the groups, the types of errors, and the interaction between these two main effects. The first source analyzes the between-subjects variability while the others analyze the within-subjects variability. Finally, naming errors were committed on 93 of the 260 assessed words. The probability to commit an error among those 93 words was compared among the groups and among the three psycholinguistic parameters (i.e., frequency, subjective frequency, and category) using a logistic regression model with the participant treated as a random effect. The addition of this random effect made it possible to take into account the dependence between a participant's observations. For all analyses, Tukey-Kramer multiple comparisons were performed on a significant result of a variation source. Analyses were performed using the SAS software for Windows (version 9.4, SAS Inc., NC) with a significance level of $p \leq .05$.

Individual's profile. Two of the authors (ML and LM) analyzed clinically the data for all participants to determine the main functional origin of anomia (i.e., lexical or semantic) based on error patterns and efficacy of semantic and phonological cueing. Specifically, for each participant, the number of each type of error was calculated as well as the percentage of efficacy of semantic and phonological cueing allowing the classification into lexical (associated with a predominance of coordinate semantic paraphasias and precise circumlocutions as well as good efficacy of phonological cueing) and semantic (associated with a predominance of non-responses, coordinate and superordinate semantic paraphasias, and vague circumlocutions along with a poor efficacy of phonological cueing) profiles. Following clinical analyses, participants ($n = 3$) with profiles not clearly related to a semantic or lexical origin were re-analyzed conjointly by the two authors to reach a consensus.

Results

The three groups did not differ significantly in age and education. There were significantly more men in the MCI group, but since results remained unchanged when sex was added as a covariate, results of the present study were not corrected for this variable. **Table 1** shows that none of the HC participants had a self-reported cognitive complaint, but more than half of the MCI participants and two-thirds of the AD patients subjectively complained about their cognitive functioning. Compared to the HC group, participants with MCI showed cognitive impairments in several domains, namely general cognitive functioning, episodic memory, executive functions, and semantic verbal fluency. In addition, there were more depressive symptoms among MCI participants than for those in the HC group. Participants with AD also had depressive symptoms as well as cognitive impairments in almost every domain except semantic memory, visual perception, and visuoconstruction. Cognitive impairments were generally more severe in the AD than in the MCI group.

Table 1

Sociodemographic and Clinical Data of the Participants

	HC (n = 15)	MCI (n = 20)	AD (n = 5)		
	M (*SD*)	*M* (*SD*)	*M* (*SD*)	*F* or χ^2	*p*
Sociodemographic characteristics					
Age (years)	70.7 (7.8)	71.8 (7.8)	77.2 (8.9)	1.3	.285
Sex (male/female)	6/9	14/6	3/2	18.4	< .001
Education (years)	14.9 (3.5)	14.5 (2.5)	15.0 (3.6)	0.1	.973
General cognitive functioning					
MoCA (/30)	26.8 (2.4)[c]	24.4 (2.5)	20.3 (2.3)[a]	11.6	**.001**
Cognitive complaint					
QPC (% of participants with a complaint)	0[bc]	53.8	66.7	8.1	**.018**
Depressive symptoms					
GDS (/30)	1.4 (2.2)[bc]	5.0 (3.3)	6.7 (5.0)	6.6	**.004**
Episodic memory					
16-item mean free recall (/16)	15.1 (1.3)	12.3 (2.4)	5.7 (2.5)	27.2	**< .001**[d]
16-item mean total recall (free + cue) (/16)	15.1 (1.1)	12.8 (2.5)	6.3 (3.1)	21.9	**< .001**[d]
16-item delayed free recall (/16)	15.7 (0.5)	13.5 (2.3)	6.7 (1.2)	29.0	**< .001**[d]
16-item delayed total recall (free + cued) (/16)	15.8 (0.4)	13.2 (2.3)	6.3 (3.1)	31.6	**< .001**[d]
Recall of the Rey Complex Figure (3 min) (/36)	19.6 (6.0)	13.3 (4.5)	0.3 (0.6)	19.9	**< .001**[d]
Semantic memory					
PPTT (/52)	50.2 (1.3)	49.5 (1.6)	49.8 (1.9)	0.9	.415
Perception/visual gnosis and visuoconstructive abilities					
Size-match task of the BORB (/30)	27.6 (1.3)	26.3 (1.9)	25.7 (2.3)	3.0	.063
Object decision (/32)	27.0 (1.8)	27.1 (2.3)	26.8 (1.7)	0.0	.992
Copy of the Rey Complex Figure (/36)	29.9 (4.4)	30.8 (4.3)	28.5 (4.0)	0.4	.649
Executive functions					
Stroop D-KEFS, Inhibition (seconds)	60.3 (10.6)	80.8 (15.2)	102.3 (6.0)	17.3	**< .001**[d]
Stroop D-KEFS, Inhibition/Shifting (seconds)	68.9 (18.0)[c]	83.4 (30.0)[c]	180.0 (0.0)[ab]	17.1	**< .001**
Stroop D-KEFS, Inhibition (errors)	1.2 (1.2)[c]	3.2 (1.8)	6.7 (9.0)[a]	5.4	**.010**
Stroop D-KEFS, Inhibition/Shifting (errors)	1.9 (1.6)	4.4 (5.4)	4.5 (2.1)	1.6	.217
Language					
Verbal fluency, letters T-P-N	38.6 (9.4)[c]	32.6 (7.1)	24.7 (6.8)[a]	4.6	**.018**
Semantic fluency, animals	18.6 (4.2)[b]	14.3 (4.8)[a]	14.0 (7.5)	3.6	.037

Note. Text in bold indicates a statistically significant difference. AD = Alzheimer's disease; BORB = Birmingham Object Recognition Battery; D-KEFS = Delis-Kaplan Executive Function System; GDS = Geriatric Depression Scale; HC = Healthy cognitive aging; MCI = Mild cognitive impairment; MoCA = Montreal Cognitive Assessment; PPTT = Pyramids and Palm Trees Test; QPC = Questionnaire de plainte cognitive; RL/RI = Free recall/cued recall 16 items.
[a]*p* < .05 compared to HC. [b]*p* < .05 compared to MCI. [c]*p* < .05 compared to AD. [d]The three groups are different *p* < .05.

Number of Errors: Differences Among the Groups

A significant difference was found among the groups for mean number of naming errors in spontaneous answers, $\chi^2(2) = 38.13$, $p < .001$. Likelihood ratios showed a significant difference among the three groups, with the AD group committing the highest mean number of errors (21.8 ± 2), followed by the MCI group (14.5 ± 0.8) and the HC (10.2 ± 0.8) group.

Errors Patterns: Differences Among the Groups

Analyses revealed no interaction effect between Group and Types of Errors, $F(22, 407) = 0.79$, $p = .734$. As a main effect, the effect of Group remained non-significant, $F(2, 37) = 0.26$, $p = .771$, while the effect of Types of Errors was significant, $F(11, 407) = 40.31$, $p < .001$. The absence of an interaction between both factors reveals a similar pattern of errors among the three groups. Therefore, the following results (mean number of errors) include all study participants. Multiple comparisons for the Types of Errors effect indicated that participants committed mostly coordinate semantic errors, followed by errors of the following types: visuosemantic (likely to be semantic since no visual agnosia was found on the Birmingham Object Recognition Battery), precise circumlocutions, non-responses, superordinate semantic, vague circumlocutions, associative categorical semantic, visual, errors classified as "others," phonological paraphasia, perseverations, and sequences of phonemic approximations (see **Table 2**). No tip-of-tongue, neologisms, or stereotypy were observed.

Efficacy of Semantic and Phonological Cueing

Analyses revealed no significant difference among the groups' percentages of correct responses following semantic, $F(2, 37) = 2.21$, $p = .124$, or phonological, $F(2, 36) = 0.11$, $p = .893$, cues. As for intragroup analysis, semantic and phonological cueing improved naming: 18.8% and 40.5% for the AD group, 16.6% and 42.0% for the MCI group, and 28.2% and 45.6% for the HC group, respectively. Given the sequential order in which the semantic and phonological cues were given, it was impossible to determine whether this intragroup difference was significant.

Influence of Psycholinguistic Parameters

The analyses revealed no interaction between Group and the three psycholinguistic parameters: Category, $F(2, 3658) = 0.32$, $p = .728$, subjective frequency, $F(4, 3658) = 1.21$, $p = .304$, and frequency, $F(4, 3658) = 1.25$, $p = .289$. As a main effect, Group remained non-significant, $F(2, 37) = 0.84$, $p = .441$, while only subjective frequency, $F(2, 3658) = 13.70$, $p < .001$, significantly influenced naming performance. Multiple comparisons showed that participants committed more errors when they had to name less familiar words (see **Table 3**). However, category, $F(1, 3658) = 0.93$, $p = .335$, and frequency, $F(2, 3658) = 0.29$, $p = .749$, did not significantly influence the probability of a naming error.

Individual Profiles

While anomia in the HC and AD groups was clearly related to lexical access difficulty (80%) and semantic deterioration (80%), respectively, two profiles were found for the MCI group. The origin of anomia was lexical for 60% of the MCI participants and semantic for 40%. When compared, the two subgroups did not differ in terms of age ($p = .235$), education ($p = .092$), or MoCA score ($p = .078$).

Discussion

The aim of this study was to document the functional origin of anomia in participants with MCI, in comparison to individuals with AD or HC, by performing a quantitative and qualitative analysis of naming errors. The efficacy of semantic and phonological cueing and the influence of psycholinguistic parameters were taken into account. Despite the relatively small number of naming errors made by all participants in the study, results showed a significant difference among the groups. Namely, participants with AD made the most errors, followed by MCI participants, and finally HC. However, the pattern of naming errors was similar in the three groups. Coordinate semantic errors, precise circumlocutions, non-responses, superordinate semantic errors, and vague circumlocutions were the most frequent types of incorrect responses. Regarding psycholinguistic parameters, results from our study showed that subjective frequency had a significant influence on the probability to commit an error in the three groups.

Quantitative and Qualitative Analysis of Naming Errors

The present study showed that individuals with AD committed more naming errors than individuals with MCI or HC, which is in line with results obtained in previous studies (Balthazar et al., 2008; Lin et al., 2014; Willers et al., 2008). Regarding participants with MCI, the present results are consistent with studies that showed the presence of anomia in individuals with MCI (Adlam et al., 2006; Balthazar et al., 2008; Balthazar et al., 2010; Dudas et al., 2005). However, one should keep in mind that other studies found no significant difference in the naming capacity of MCI and HC participants (Beinhoff et al., 2005; Willers et al., 2008). Despite a significant difference among the groups, AD and MCI participants committed only a few naming errors, thus suggesting the presence of naming difficulties rather than a naming impairment.

Table 2

Total Number of Each Type of Naming Error Committed by all Participants

	All participants (*n* = 40)	
Type of Naming Error	Total (*SD*)	Tukey-Kramer[a]
Coordinate semantic paraphasias	42.3 (2.9)	a
Visuosemantic paraphasias	11.9 (1.5)	b
Precise circumlocutions	10.9 (1.4)	b
Non-responses	8.9 (1.3)	cb
Superordinate semantic paraphasias	8.5 (1.2)	cb
Vague circumlocutions	6.3 (1.1)	bcd
Associative semantic paraphasias	4.7 (0.9)	cde
Visual	3.4 (0.8)	de
Others	2.2 (0.6)	de
Phonological paraphasias	0.5 (0.3)	e
Perseverations	0.2 (0.2)	e
Sequences of phonemic approximations	0.2 (0.2)	e

Note. [a]Comparisons sharing a same letter are not significantly different.

Table 3

Naming Errors Committed by all Participants According to Psycholinguistic Parameters

	All participants (*n* = 40)		
	M (*SD*)	*F*	*p*
Subjective frequency		13.3	< .001
High	9.7 (2.5)		
Moderate	14.5 (3.7)		
Low	28.8 (7.2)		
Frequency		1.2	.315
High	9.5 (6.5)		
Moderate	23.1 (4.6)		
Low	19.0 (2.0)		
Category		1.0	.327
Biological	17.2 (4.2)		
Manufactured	15.5 (3.8)		

The similar pattern of naming errors among the three groups in the present study is also consistent with results from previous studies (i.e., Balthazar et al., 2008; Lin et al., 2014; Willers et al., 2008). More specifically, Balthazar et al. (2008) showed a similar pattern of naming errors among HC, MCI, and AD participants. Lin et al. (2014) reported a similar distribution of the types of errors in individuals with AD and controls, but participants with MCI were not included in the study. Finally, Willers et al. (2008) found a similar pattern of errors in participants with AD and MCI, but not in controls who committed significantly less semantic errors. Taking into account only the type of naming errors, our results suggest an implication of the same components in naming in individuals with AD, MCI, and HC, with a gradual degradation of these components following the severity of the cognitive impairment.

Regarding the types of errors found in the present study, there are many similarities with data from previous work. Two main findings can be drawn: (a) coordinate semantic errors are very common, while (b) phonological paraphasias are very rarely produced by HC, MCI, or AD participants. The strong presence of coordinate semantic paraphasias could suggest an impairment in the access or retrieval of the semantic features among the semantic memory, in line with the literature targeting a semantic origin for anomia in MCI (Adlam et al., 2006; Joubert et al., 2010; Willers et al., 2008), but also could suggest a lexical access difficulty (Whitwort et al., 2013), targeting a more lexical origin. Since precise circumlocutions, usually associated with a lexical origin, were the second most frequent type of errors, this must be interpreted carefully. Moreover, results from the present study allowed us to be more specific about the types of errors reported in the literature. Indeed, in this study, compensation strategies for anomia like circumlocutions were differentiated into precise circumlocutions, habitually associated with a lexical impairment (i.e., difficulty to access the precise word but spared semantic knowledge) and vague circumlocutions, rather associated with a semantic deficit (i.e., difficulty to access the word and impaired semantic knowledge), a distinction that was not made in previous work. In this study, precise circumlocutions were more frequent than vague circumlocutions.

Psycholinguistic Parameters and Cueing

For all participants, errors were mostly committed for less familiar words, without significant effects of category and frequency. The absence of category and frequency effects could be explained by a lack of statistical power caused by high variability in the parameters associated with the 93 words that yielded an error at least once in the naming task. A bigger sample would have allowed us to observe higher numbers of naming errors, thus increasing statistical power. Another explanation could be that the analysis was conducted on all participants regarding psycholinguistic parameters despite some having anomia related to lexical access difficulty (80% of HC and 60% of MCI), which is usually associated with an impact of the frequency, while the rest had anomia related to semantic deterioration (80% of AD and 40% of MCI), which is usually associated with an impact of other psycholinguistic parameters such as category. The absence of a category effect contrasts with the results of some studies. For example, several authors reported greater difficulties in naming for biological objects compared to manufactured ones in individuals with AD or MCI (Callahan et al., 2015; Duong et al., 2006; Fung et al., 2001; Taler et al., 2016; Whatmough et al., 2003). However, other studies also have revealed the absence of a category effect on naming, as in the current study (Laws et al., 2007; Lockyer, Sheppard, & Taler, 2015).

The efficacy of phonological cueing to facilitate naming in the three groups is consistent with previous studies (Balthazar et al., 2008; Willers et al., 2008) and could suggest a lexical origin for anomia. However, it was impossible to determine if the cues had a significant influence on naming performance.

Functional Origin of Anomia

The cognitive model proposed by Caramazza and Hillis (1990) suggests that the type of naming errors is determined by the functional origin of the impairment. When all participants were considered, the error patterns, the efficacy of cueing, and the influence of psycholinguistic parameters did not point in a clear direction to the functional origin of anomia since manifestations of both lexical and semantic impairment were found. Therefore, a separate individual analysis was conducted for each participant to see if the unclear group profile could result from the combination of two distinct individual profiles, namely lexical and semantic. This hypothesis was confirmed by the individual analyses. For some participants, the semantic system seemed to be the main cause of anomia (n = 15), while for the others, the phonological output lexicon and its access appeared as the principal cause (n = 25). More interestingly, the main origin of anomia varied among the three groups. For HC participants, the origin of the observed anomia was mostly lexical (80%), while a more combined lexico-semantic origin was identified as the possible cause of anomia for three HC participants. This result is in line with literature showing that semantic knowledge is well preserved in healthy aging (Piolino, Desgranges, Benali, & Eustache, 2002). In the MCI group in the present study, results were mixed with anomia being caused by an impairment of lexical access for most of the participants (60%) and by an impairment of the semantic system for 40%. In the literature, results were also inconsistent, with some studies suggesting a semantic origin (Adlam et al., 2006; Joubert et al., 2010; Willers et al., 2008), and others a lexico-semantic origin (Balthazar et al., 2008; Duong et al., 2006; Guidi et al., 2015; Taler & Phillips, 2008) of anomia in people with MCI.

According to the individual analyses performed in the current study, this inconsistency found in previous work could be explained by the presence of two distinct profiles. In this study, participants in the two subgroups (i.e., lexical impairment/semantic impairment) did not differ in age, education, and general cognitive functioning assessed by the MoCA. Thus, age, level of education, and general cognitive functioning could not predict the profile found for a participant. Future studies should focus on understanding the differences between those two subgroups with regard to other cognitive functions, evolution, and prognosis in order to allow for more effective and accurate interventions at all stages of the disease. Finally, in the AD group, an impairment of the semantic system was identified as the main cause of anomia for 80% of the participants, which is in line with the study by Lin et al. (2014) in which a progressive degradation of the semantic system was responsible for anomia in AD. However, it is noteworthy that for the participants in the present study, the succinct objective assessment of the semantic system (with the Pyramids and Palm Trees Test) showed no semantic deficit. The Pyramids and Palm Trees Test did not seem to be sensitive enough to objectify difficulties in the access or retrieval of fine and distinctive semantic features that could be at the origin of anomia in the AD group as suggested by Lin et al. However, these results must be interpreted carefully given the very small AD sample in this study.

Nevertheless, the idea that anomia is caused by difficulties to access or retrieve the distinctive semantic features within the semantic memory is well documented in AD. Indeed, according to many researchers, distinctive semantic features would be more vulnerable to pathology than general semantic features shared by several concepts (Catricalà et al., 2015; Flanagan, Copland, van Hees, Byrne, & Angwin, 2016; Garrard, Lambon Ralph, Patterson, Pratt, & Hodges, 2005; Laisney et al., 2011). For this population, the primacy of coordinate semantic paraphasias could therefore be explained by a gradual deterioration of these distinctive semantic features, thus resulting in a difficulty to differentiate two close concepts belonging to the same category because of unclear semantic representations (Catricalà et al., 2015; Garrard et al., 2005).

Strengths and Limitations

The present work has some limitations, the most important being the small number of participants, especially for AD. Even though statistical analyses accounted for these small sample sizes, the results regarding the origin of anomia must be interpreted carefully. Moreover, the small samples may have hidden differences among the three groups regarding sociodemographic variables. Similarly, even though the oral naming task had 260 images, participants made errors on only 98 of them, lowering the statistical power of the study regarding psycholinguistic parameters. Therefore, it is crucial that those results are replicated with larger sample sizes, but this study is yet an important step towards the understanding of anomia in pathological aging. Also, participants included in the study had a mean education of 15 years, which is not representative of the older adult population. This may have influenced the preservation of their naming capacities (Reis, Guerreiro, & Castro-Caldas, 1994).

Finally, it is likely that the semantic cues were not precise enough to help naming in the case of an anomia caused by difficulties in access or retrieval of specific and distinctive semantic features. Even though the semantic cues were created systematically and administered rigorously (all participants received the exactly same cues in the same order and the same moment), they were about semantic features most commonly associated with the target word, which leads in some cases to a lack of precision to help naming. For example, for the word *apple,* the semantic cue was "red fruit." Although suitable, this cue also corresponds to the target words cherry, strawberry, and raspberry. A more specific semantic cue could have been beneficial, such as "red fruit picked from the trees in autumn." Another possibility could have been to propose to all participants a semantic survey targeting the failed items.

This study also has significant strengths. First, the exclusion of participants with visual agnosia was a significant strength, despite its impact on the sample size and statistical power. Indeed, the qualitative analysis of errors usually makes it impossible to determine whether the origin of visuosemantic errors is visual or semantic. By excluding participants with difficulties in visual recognition, we can hypothesize that their origin was generally semantic. Moreover, the exclusion of participants with probable visuoperceptual difficulties is fundamental when the experimental task uses images. In the same line, the assessment of the semantic system with the Pyramids and Palm Trees Test allowed a more nuanced interpretation of the naming performance, as explained earlier. Second, many factors allowed us to minimize the risk of errors or bias in the qualitative analysis of the results, such as the use of an exhaustive classification of naming errors, the rigorous selection of semantic and phonological cues, the standardized administration of the task, and the use of audio recordings to analyze more finely the types of errors.

Clinical Implications and Future Perspectives

The clinical relevance of the present study is based on two main findings: (a) individuals with MCI commit relatively few naming errors despite the fact that anomia is a core element of the cognitive complaint and (b) naming difficulties found in HC and anomia observed in MCI and AD has a different origin, and two distinct profiles are found in MCI. By specifying the functional origin of anomia in MCI and AD, this study is the preliminary step in developing cognitive interventions specifically targeting the naming difficulties of these populations. In MCI, the present results highlight the importance of an appropriate individual assessment prior to intervention in order to determine the functional origin of anomia since distinct profiles (i.e., lexical or semantic or a combination of lexico-semantic) can be found. In our study, the Pyramids and Palm Trees Test, which assesses semantic processing for functional and encyclopedic knowledge of the target words, did not seem to be sensitive enough to objectify difficulties in the access or retrieval of fine and distinctive semantic features, yet observed in participants with AD and 40% of participants with MCI. Therefore, the development of a more extensive and comprehensive battery to assess semantic memory would be extremely relevant, especially since no battery is actually adapted to the French-Québec population.

In addition, interventions aimed at reducing anomia in AD should target the impaired access to semantic knowledge or the compensation of the degradation of this knowledge in the semantic system. Furthermore, even though the exclusion of patients with visuospatial impairments was a strength of the study, considering that it is usual to have visuospatial impairments within AD, it would be important to conduct studies aiming to describe the specific needs of such patients in terms of language or anomia rehabilitation. Moreover, it would be important to develop other tests to evaluate the semantic system without using a pictographic entry. Until now, cognitive interventions developed for individuals with MCI mainly focused on deficits of episodic memory (Jean, Bergeron, Thivierge, & Simard, 2010; Reijnders, van Heugten, & van Boxtel, 2013; Simon, Yokomizo, & Bottino, 2012), working memory (Hyer et al., 2016), and executive functions (Mowszowski, Lampit, Walton, & Naismith, 2016). An intervention specifically targeting word-finding difficulties could potentially slow the worsening of this deficit during the decline that leads an individual with MCI to the dementia phase of AD.

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Développement, validation et normalisation de la *Batterie d'évaluation de la compréhension syntaxique* : une collaboration Québec-Suisse

Development, Validation, and Standardization of the *Batterie d'évaluation de la compréhension syntaxique*: A Québec–Switzerland Collaboration

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MOTS-CLÉS
SYNTAXE
ÉVALUATION
APHASIE
NORMALISATION

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Abrégé

Afin de combler le manque d'outils d'évaluation disponibles en français, la *Batterie d'évaluation de la compréhension syntaxique* a été élaborée conjointement par des équipes de recherche de la Suisse et du Québec. D'abord, des participants souffrant d'aphasie (n = 30) ont été recrutés afin d'évaluer les validités convergente, divergente et discriminante, ainsi que pour déterminer la fidélité test-retest de l'outil. Les résultats de ces analyses ont montré que la batterie avait une bonne validité convergente, une excellente validité divergente et une excellente fidélité test-retest. Les analyses effectuées pour déterminer la validité discriminante ont quant à elles révélé que la *Batterie d'évaluation de la compréhension syntaxique* était aussi spécifique que la tâche d'évaluation de compréhension de phrases du *Protocole Montréal-Toulouse d'examen linguistique de l'aphasie*, en plus d'être plus sensible que ce test couramment utilisé pour identifier les patients ayant été victimes d'un accident vasculaire cérébral et présentant des déficits de compréhension syntaxique. Ensuite, deux groupes de participants sains provenant de la Suisse (n = 75) et du Québec (n = 25) ont été recrutés afin d'effectuer une normalisation de la batterie. Des analyses ont été effectuées afin de comparer l'impact de la provenance géographique, de l'âge et du niveau de scolarité sur la performance à la batterie. Les résultats montrent que les participants provenant du Québec et de la Suisse performent de manière similaire. Seul le niveau de scolarité a un effet significatif sur les performances aux tâches; les normes présentées sont donc stratifiées en ce sens. Les résultats suggèrent que la *Batterie d'évaluation de la compréhension syntaxique* a de bonnes qualités métrologiques et que ses normes peuvent être utilisées auprès des populations franco-suisse et franco-québécoise.

Abstract

To fill the lack of French evaluation tools, the *Batterie d'évaluation de la compréhension syntaxique* [a battery for the evaluation of syntax processing] was developed in collaboration with research teams from Switzerland and the province of Québec. First, participants with aphasia (n = 30) were recruited to complete convergent, divergent, and discriminant validity and reliability assessments. The results showed good convergent validity, excellent divergent validity, and excellent reliability for the battery. Discriminant validity analyses revealed that the *Batterie d'évaluation de la compréhension syntaxique* is as specific as the syntactic comprehension task of the *Protocole Montréal-Toulouse d'examen linguistique de l'aphasie* and is more sensitive than this commonly used test in identifying post-stroke patients with syntactic comprehension deficits. Second, two groups of healthy participants from Switzerland (n = 75) and Québec (n = 25) were recruited to provide standardized data. Analyses were conducted to compare the impact of geographic origin, age, and education on performance on the battery. Control participants from Québec and Switzerland performed similarly on the battery. Only education level had a significant effect on performance; normative data was therefore stratified accordingly. Results showed that the *Batterie d'évaluation de la compréhension syntaxique* had good metrological qualities and norms can be used with both Québec-French and Swiss-French populations.

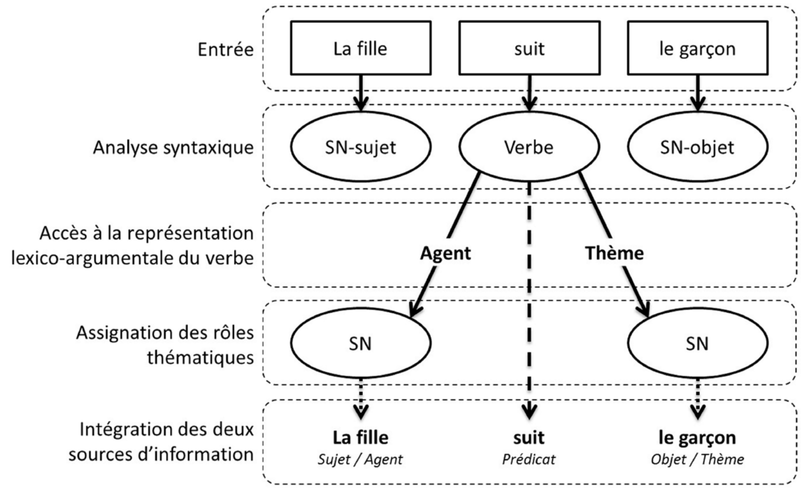
Chaque année, environ le tiers des personnes victimes d'un accident vasculaire cérébral (AVC) souffrent d'aphasie (Engelter et al., 2006). Parmi ces personnes, un grand nombre présente des difficultés de compréhension orale, pouvant se manifester par des difficultés à comprendre les mots isolés, ou même, les phrases. Dans ces cas, c'est la compréhension de la signification des phrases qui pose problème, et ce, surtout lorsque l'absence d'indices sémantiques ou contextuels contraint la personne à interpréter la phrase en ne se basant que sur sa connaissance des règles syntaxiques (Caramazza et Zurif, 1976). Ce déficit, communément appelé compréhension agrammatique ou asyntaxique (Caramazza, Berndt, Basili et Koller, 1981), renvoie donc à une difficulté à exploiter les structures syntaxiques pour déterminer le sens des phrases (Caplan, Waters, DeDe, Michaud et Reddy, 2007). Il est notamment observé chez les personnes ayant une aphasie de Broca (Rigalleau, Baudiffier et Caplan, 2004; Rigalleau, Nespoulous et Gaonac'h, 1997) ou chez les personnes atteintes de certaines variantes de l'aphasie primaire progressive (p. ex. la variante agrammatique non-fluente; Charles et al., 2014; Gorno-Tempini et al., 2011).

Les processus de compréhension syntaxique sont complexes et impliquent plusieurs niveaux de traitement, qui sont eux-mêmes influencés par des processus cognitifs tels que l'attention ou les fonctions exécutives (Key-DeLyria et Altmann, 2016). Déterminer la nature du déficit sous-jacent aux difficultés de compréhension syntaxique est donc une tâche ardue, d'autant plus que différentes conceptions théoriques coexistent dans la littérature. Parmi les modèles disponibles pour rendre compte des processus de compréhension de phrases, celui de Saffran, Schwartz, Fink, Myers et Martin (1992) est particulièrement intéressant, car il repose sur des étapes distinctes de traitement, facilement transposables en épreuves d'évaluation clinique.

La figure 1 présente le modèle de compréhension de phrases de Saffran et al. (1992), composé de quatre étapes de traitement. Ce modèle a été largement étudié dans la littérature et a été utilisé comme base théorique dans l'élaboration de certains tests de compréhension syntaxique en anglais, notamment dans l'élaboration du *Verb and Sentence Test* (Bastiaanse, Edwards et Rispens, 2002). La première étape correspond à l'analyse syntaxique (ou *parsing*), qui consiste à analyser syntaxiquement la séquence de mots (en l'occurrence, la phrase). En d'autres mots, cette étape sert à déterminer le type de syntagme des constituants de la phrase (p. ex. syntagme nominal, syntagme verbal, syntagme adjectival) et leur position ou fonction syntaxique (c.-à-d. position sujet, position verbe, position objet). La seconde étape de traitement correspond à l'accès à la représentation lexico-argumentale du verbe. Celle-ci vise à récupérer les arguments du verbe (selon les verbes, ceux-ci peuvent être au nombre de 1, de 2 ou de 3) et les rôles thématiques (le « qui fait quoi à qui ») qui leur sont associés et qui sont encodés dans l'entrée lexicale du verbe. Comme la signification du verbe ne peut être dissociée de ses arguments et de leurs rôles thématiques, cette étape implique donc de d'accéder à la signification du verbe. La troisième étape correspond à l'assignation des rôles thématiques (ou *mapping*). Elle consiste à assigner les rôles thématiques du verbe (définis à l'étape 2) aux positions syntaxiques (définies à l'étape 1). Lorsque les constituants de la phrase respectent l'ordre canonique (sujet – verbe – objet, pour une langue comme le français), l'assignation des rôles thématiques est relativement simple puisque le sujet est habituellement l'agent de l'action et l'objet est le thème (p. ex. la fille$_{\text{sujet/agent}}$ pousse$_{\text{verbe}}$ le garçon$_{\text{objet/thème}}$). Cependant, dans les phrases qui ne respectent pas l'ordre canonique des constituants (comme les phrases passives ou les phrases subordonnées objet), l'assignation des rôles thématiques est plus délicate à faire. En effet, comme les constituants ont été déplacés, le sujet ne joue plus le rôle d'agent, mais joue le rôle de thème et l'objet ne joue plus le rôle de thème, mais celui d'agent (p. ex. le garçon$_{\text{sujet/thème}}$ est poussé$_{\text{verbe}}$ par la fille$_{\text{objet/agent}}$). Finalement, la quatrième et dernière étape du modèle renvoie à l'activation de la représentation complète du sens de la phrase. Cette dernière étape est combinée avec la troisième, car elle n'est pas opérationalisable de manière isolée (Schwartz, Saffran, Fink, Myers et Martind, 1994).

Bien que beaucoup d'importance ait été accordée aux différentes étapes du modèle de Saffran et al. (1992) dans l'étude des processus de compréhension de phrases concernant les problématiques associées à l'aphasie (Caplan et al., 2007; Schwartz et al., 1994; Shapiro et Levine, 1990; Wilson et Saygın, 2004), force est de constater qu'elles ont rarement été prises en compte dans le développement des épreuves d'évaluation disponibles en clinique. À notre connaissance, il n'existe aucun instrument d'évaluation en français explorant spécifiquement chacune de ces étapes. L'épreuve de compréhension de phrases de la version β du *Protocole Montréal-Toulouse d'examen linguistique de l'aphasie* (MT-86; Nespoulous et al., 1992) demeure l'un des principaux tests utilisés en clinique auprès de la clientèle adulte aphasique francophone. Toutefois, cette épreuve permet seulement d'évaluer la compréhension des phrases globalement (c.-à-d. la dernière étape du modèle de Saffran et al., 1992); elle ne permet de tester ni l'étape d'analyse syntaxique (ou *parsing*) ni l'étape d'accès aux représentations lexico-argumentales du verbe. Cette

Figure 1



Adaptation du modèle de compréhension de phrases de Saffran, Schwartz, Fink, Myers et Martin (1992), tel que décrit dans Caron, Le May, Bergeron, Bourgeois et Fossard (2015). SN = syntagme nominal.

épreuve ne permet donc pas d'identifier précisément à quelle étape du modèle de Saffran et al. se situerait le déficit de compréhension de phrases d'une personne. Une autre lacune de ce protocole est l'absence de contrôle des paramètres psycholinguistiques maintenant reconnus pour influencer les étapes du traitement syntaxique (p. ex. la fréquence des mots, le nombre d'arguments des verbes, la complexité syntaxique, la longueur des phrases).

Récemment, deux outils d'évaluation informatisés de la compréhension syntaxique ont été publiés en français, soit le *Test informatisé de compréhension syntaxique en français* (Python, Bischof, Probst et Laganaro, 2012) et le sous-test de compréhension de phrases de l'outil *GREMOTs : batterie d'évaluation des troubles du langage dans les maladies neurodégénératives* (Bézy, Renard et Pariente, 2016). Ces deux tests, standardisés et normés auprès d'une population adulte francophone, présentent de nombreux avantages, dont un meilleur contrôle des paramètres psycholinguistiques d'intérêt (c.-à-d. fréquence et diversité lexicale, diversité des structures syntaxiques testées, contrôle des distracteurs). Toutefois, comme pour l'épreuve du MT-86, ces deux tests n'évaluent que la phase finale des processus de compréhension de phrases (c.-à-d. l'étape d'assignation des rôles thématiques ou *mapping*), ce qui ne permet pas de localiser l'origine fonctionnelle probable du déficit.

D'autres tests mettant à contribution l'état actuel des connaissances sur la syntaxe existent dans d'autres langues, en particulier en anglais, tels que le *Verb and Sentence Test* (Bastiaanse et al., 2002) et le *Northwestern Assessment of Verbs and Sentences* (Thompson, 2012). Outre le fait que ces tests n'évaluent pas l'ensemble des étapes de la compréhension de phrases, leur traduction et adaptation en français ne sont néanmoins pas sans poser problème, surtout lorsqu'on considère les paramètres psycholinguistiques qui diffèrent d'une langue à une autre. En conséquence, il est généralement difficile d'évaluer de façon objective et avec des tests basés sur des théories actuelles les troubles de compréhension syntaxique des personnes aphasiques francophones. Pourtant, les difficultés de compréhension de phrases méritent d'être cliniquement bien investiguées, car elles peuvent sévèrement limiter l'étendue des significations auxquelles une personne aphasique peut accéder. Une évaluation plus précise permet donc de mieux cerner les profils de communication au plan réceptif, en plus de permettre de mettre en place des programmes de rééducation plus spécifiques.

Afin de répondre à ce besoin clinique et de combler les lacunes identifiées avec les instruments existants, une batterie d'évaluation de la compréhension syntaxique a été élaborée, soit la *Batterie d'évaluation de la compréhension syntaxique* (BCS; Caron, Le May et Fossard, 2010). Celle-ci est basée sur le modèle théorique de Saffran et al. (1992), composé de quatre étapes, et est le fruit d'une collaboration entre des équipes de recherche de la Suisse et du Québec. Les objectifs du présent article sont de présenter les trois phases ayant mené à l'élaboration complète de l'outil : le développement (phase 1), la validation (phase 2) et la normalisation (phase 3) de la BCS.

Phase 1 : développement de l'outil

L'objectif de cette première phase était de créer l'outil et d'en valider le contenu. Une première version de la BCS (simplifiée BECS à l'époque) a donc été créée en 2010 par des étudiantes inscrites dans le programme de maîtrise en orthophonie à l'Université Laval à Québec, sous la direction de Marion Fossard. À partir d'une évaluation approfondie des outils de mesure existants et s'appuyant sur les données de la littérature, cinq épreuves originales destinées à tester les étapes du modèle de compréhension de phrases de Saffran et al. (1992) ont été créées.

La première étape du modèle, soit l'étape d'analyse syntaxique (ou *parsing*), est évaluée au moyen de deux épreuves (tâches A-1 et A-2). L'épreuve de catégorie grammaticale (tâche A-1) permet de vérifier si une personne peut accéder à la catégorie grammaticale des mots de son lexique en distinguant un nom (p. ex : livre, chasseur) d'un verbe (p. ex : lire, chasser), et ce, sur la base d'une analyse morphologique. Cette habileté est en effet indispensable au découpage d'une phrase en syntagmes lors de l'analyse syntaxique. L'épreuve de jugement de grammaticalité (tâche A-2) évalue la capacité d'une personne à construire une représentation syntaxique de la phrase (selon un jugement sur la syntaxe) et teste donc l'analyse syntaxique (ou *parsing*). La deuxième étape du modèle, soit l'accès à la représentation lexico-argumentale du verbe, est évaluée via une épreuve de représentation lexico-argumentale du verbe (tâche B). Cette tâche permet d'évaluer précisément la capacité d'une personne à récupérer l'information relative au nombre d'arguments d'un verbe. Finalement, la dernière étape du modèle, soit l'étape d'assignation des rôles thématiques et d'intégration des sources d'information (ou *mapping*), est évaluée au moyen de deux épreuves (tâches C-1 et C-2). La tâche C-1 est un prétest de compréhension lexicale qui vise simplement à confirmer la capacité d'une personne à accéder au sens des concepts utilisés dans la tâche C-2. L'épreuve d'assignation des rôles thématiques (tâche C-2) vérifie, dans diverses configurations syntaxiques, si une personne est capable d'assigner correctement les rôles thématiques aux bons constituants d'une phrase (Caron, Le May, Bergeron, Bourgeois et Fossard, 2015).

À la suite de cette première version, une étude de validité de contenu a été réalisée (Bergeron, Bourgeois, Fossard, Desmarais et Lepage, 2013). Un questionnaire de validation de contenu a d'abord été développé par l'équipe de recherche. Le questionnaire utilisait des échelles de Likert composées de quatre niveaux allant de « tout à fait d'accord » à « tout à fait en désaccord » et permettait d'analyser les différentes composantes de la BCS (c.-à-d. les explications théoriques, le choix des items-test et des items d'entrainement, la clarté des consignes, la clarté des images utilisées, la notation des résultats, la grille d'analyse, ainsi que l'utilité/l'apport clinique de la BCS).

Douze experts (c.-à-d. trois chercheurs dans le domaine de l'aphasie et de la syntaxe et neuf orthophonistes cliniciens) ont été sollicités pour compléter le questionnaire. Afin de s'appuyer sur une utilisation pratique de la BCS, les orthophonistes cliniciens ont administré la batterie à 24 personnes aphasiques au total, recrutés parmi leurs propres patients, avant de compléter le questionnaire de validation de contenu. Les données recueillies via le questionnaire ont ensuite été soumises à une analyse quantitative et qualitative. Les résultats obtenus ont permis de montrer la validité de contenu de la batterie avec un accord variant entre 83% et 93% selon les sections et les aspects évalués. Des éléments à améliorer ont également été identifiés. Ceux-ci concernaient par exemple l'ambiguité de certaines images, les explications théoriques, le choix de certains items-test, la formulation des consignes et la configuration des grilles de notation du test. À la suite du processus de validation de contenu, l'équipe de recherche a effectué des modifications à l'outil initial afin d'en arriver à une version améliorée. La version finale de la BCS a été publiée en 2015 par le Centre intégré universitaire de santé et services sociaux de la Capitale-Nationale – Institut de réadaptation en déficience physique de Québec (CIUSSS de la Capitale-Nationale – IRDPQ) et est disponible à l'adresse www.ciussscn.ca/BCS.

Le **tableau 1** présente le détail de chacune des tâches de la batterie : les types de stimulis utilisés, l'étape à laquelle elle réfère dans le modèle théorique, de même que les paramètres psycho-linguistiques contrôlés. La durée d'administration de la version finale de la batterie est d'environ 60 minutes et ne nécessite comme matériel que le cahier de dessins et les formulaires de réponse. Un guide du clinicien accompagne aussi l'outil.

Phase 2 : validation de l'outil

La seconde phase du projet avait pour objectif de valider la BCS en évaluant ses qualités métrologiques. La validité de construit de l'outil a été établie en investiguant la validité convergente, la validité divergente et la validité discriminante. La fidélité test-retest a également été évaluée.

Participants et procédure de recrutement

Afin d'établir la validité de construit et la fidélité test-retest, 30 personnes devenues aphasiques à la suite d'un AVC ont été recrutées parmi les patients, actifs ou anciens, du CIUSSS de la Capitale-Nationale – IRDPQ.

Tableau 1

Descriptif des épreuves de la *Batterie d'évaluation de la compréhension syntaxique*

Étape du modèle théorique[1]	Nom de la tâche	Type de tâche	Matériel	Paramètres psycholinguistiques contrôlés
Analyse syntaxique	A-1 : Catégorie grammaticale	Appariement mot entendu/ image	4 items d'entrainement 20 items-test	Longueur et fréquence des stimuli Position de la cible sur la planche
	A-2 : Jugement de grammaticalité	Jugement de grammaticalité	3 items d'entrainement 36 items-test	Complexité syntaxique et grammaticalité des phrases
Accès à la représentation lexico-argumentale du verbe	B : Représentation lexico-argumentale du verbe	Appariement mot entendu/ image	2 items d'entrainement 10 items-test	Fréquence des stimuli Nombre d'argument des verbes Position de la cible sur la planche
Assignation des rôles thématiques et intégration des deux sources d'information	C-1 : Pré-test de compréhension lexicale	Appariement mot entendu/ image	10 items-test	Position de la cible sur la planche
	C-2 : Assignation des rôles thématiques	Appariement phrase entendue/ image	2 items d'entrainement 50 items-test répartis en 2 blocs	Type de structure syntaxique Choix des adjectifs Structure argumentale des verbes Longueur des stimuli Fréquence des verbes Caractéristiques du sujet (agent) - objet (thème) Position du sujet Position de la cible sur la planche

Note. [1]Selon Saffran, Schwartz, Fink, Myers et Martin (1992).

Ces participants avaient tous le français comme langue maternelle (principale langue parlée au domicile durant l'enfance et au moment de l'étude), étaient âgés entre 21 et 90 ans (M = 65,3; $ÉT$ = 15,4) et présentaient un niveau de scolarité variant entre 4 et 16 ans (M = 11,0 , $ÉT$ = 3,2). Parmi ces participants, on comptait 17 hommes et 13 femmes. Sur les 30 participants, 17 avaient subi un AVC dans les 6 derniers mois et étaient toujours en réadaptation active, tandis que l'AVC des 13 autres était survenu plus de 6 mois avant le début de l'étude (entre 6 et 168 mois, M = 45). Ces 13 participants n'étaient donc plus suivis en réadaptation intensive. Les participants ont été recrutés selon les critères d'inclusion suivants : 1) avoir une conclusion orthophonique d'aphasie post-AVC, 2) avoir 18 ans et plus et 3) avoir le français comme langue maternelle. Les critères d'exclusion étaient : 1) présenter un déficit visuel ou auditif important non corrigé, 2) avoir un diagnostic actuel d'épisode dépressif majeur (critères de la 5e édition du *Manuel diagnostique et statistique des troubles mentaux*; American Psychiatric Association, 2016) et 3) avoir des capacités cognitives et motrices ne permettant pas à la personne d'exécuter l'ensemble des tâches contenues dans la BCS.

Procédures de collecte de données

Le projet a été approuvé par le comité d'éthique de la recherche de l'Institut de réadaptation en déficience physique de Québec (certificat d'éthique #2015-450). Cinq orthophonistes du programme des encéphalopathies du CIUSSS de la Capitale-Nationale – IRDPQ et deux stagiaires d'internat en orthophonie ont administré les tests, après avoir reçu une formation leur permettant d'assurer une passation standardisée. La cueillette de données s'est échelonnée sur 21 mois au total. Une fois le consentement signé par le participant, chaque orthophoniste devait administrer à son patient la BCS au complet (5 épreuves), ainsi que la tâche de compréhension orale de mots et de phrases du MT-86 (Nespoulous et al., 1992) et la tâche de prosodie réceptive émotionnelle du *Protocole Montréal d'évaluation de la communication* (MEC; Joanette, Ska et Côté, 2004). La durée totale de passation de l'ensemble des tâches variait entre une et deux heures et l'orthophoniste pouvait répartir ce temps en deux séances au besoin (comme cela est normalement fait en clinique). Neuf participants dont l'AVC datait de plus de 6 mois (aphasie chronique) ont complété une deuxième fois la BCS, 3 mois après la première passation (M = 4,7 mois), afin d'évaluer la fidélité test-retest de l'instrument.

Outils de mesure et analyses

Afin de mesurer la validité convergente, les résultats obtenus à la tâche C-2 de la BCS ont été comparés, à l'aide de tests de corrélation de Pearson, à ceux obtenus à la tâche d'évaluation de compréhension de phrases du MT-86 (Nespoulous et al., 1992). Dans cette épreuve du MT-86, le participant doit pointer une image en réponse à une phrase entendue, et ce, parmi un choix composé de deux, quatre ou six images. Cette épreuve a été choisie pour l'étude de la validité convergente car, tout comme la tâche C-2 de la BCS, elle évalue la compréhension syntaxique. Par exemple, le MT-86 propose des phrases simples ou complexes, réversibles ou non, telles que « C'est le garçon que le chien regarde. » ou encore « Le cheval tire le garçon. » Les différentes structures de phrases sont reprises plusieurs fois au cours du test et le participant doit pointer parmi plusieurs images celle qui correspond le mieux à la phrase entendue. Une corrélation de plus de 0,70 était attendue pour montrer la validité convergente de l'outil.

Afin de mesurer la validité divergente, les résultats obtenus à la tâche C-2 de la BCS ont été comparés, à l'aide de tests de corrélation de Pearson, à ceux obtenus dans la tâche de compréhension de la prosodie émotionnelle du MEC (Joanette et al., 2004). Dans cette épreuve du MEC, le participant entend des phrases préenregistrées et doit pointer parmi trois choix (colère, joie ou tristesse) l'émotion correspondant à l'intonation entendue sur l'enregistrement. Cette épreuve a été choisie pour évaluer la validité divergente parce qu'elle évalue elle aussi la compréhension, mais n'implique aucune compréhension lexicale ou syntaxique. Une absence de corrélation significative entre les résultats des deux tâches était attendue pour montrer la validité divergente de l'outil.

Dans le contexte clinique, la validité discriminante réfère au potentiel d'un outil à identifier adéquatement, parmi un groupe d'individus, les personnes ayant des troubles de ceux qui n'en ont pas. La validité discriminante de la BCS a été mesurée en comparant les participants identifiés comme ayant un trouble de compréhension syntaxique à la BCS (somme des résultats aux tâches C-1 et C-2) à ceux identifiés comme ayant un trouble de compréhension syntaxique au MT-86 (tâche de compréhension orale de mots et de phrases). Cette comparaison a été mesurée avec l'aide d'un test de chi-carré de Pearson. À noter que cette identification de participants s'est basée sur les données normatives publiées pour le MT-86 (Béland et Lecours, 1990) où les scores critiques (< 5^{e} percentile) varient entre 36/47 et 40/47 selon l'âge et le niveau de scolarité des participants. Pour la BCS, cette identification (< 5^{e} percentile) s'est basée sur les données normatives présentées dans le présent article à la section « Phase 3 : normalisation de l'outil » (voir le **tableau 3**). Pour les fins de cette analyse, une valeur normative a été calculée pour le score combiné des tâches C1 et C2.

Afin de mesurer la fidélité test-retest, les résultats des 9 participants ayant une aphasie chronique et ayant complété la BCS une deuxième fois au moins 3 mois après la première passation ont été étudiés. Une analyse de coefficient de corrélation intra-classe à modèle mixte à deux facteurs de type cohérence absolue a été effectuée sur les résultats aux cinq tâches de la BCS. Une corrélation intra-classe de plus de 0,75 suggère une fidélité test-retest adéquate. Ces analyses suivent les recommandations de Koo et Li (2016).

Résultats

Pour l'établissement de la validité convergente, une analyse de corrélation de Pearson a été effectuée entre le score à la tâche C-2 de la BCS et le score à la tâche de compréhension de phrases du MT-86. Une corrélation positive significative a été obtenue entre les deux scores, $r(28) = 0{,}705$, $p < 0{,}001$. Ce résultat indique que la performance aux tâches de compréhension de phrases des deux tests évaluant un construit similaire est fortement corrélée et que la validité convergente de la BCS est bonne.

Pour l'établissement de la validité divergente, une analyse de corrélation de Pearson a été effectuée entre le score à la tâche C-2 de la BCS et le score à la tâche de compréhension prosodique du MEC. Aucune corrélation significative n'a été obtenue entre les deux scores, $r(28) = 0{,}146$, $p = 0{,}443$. Ce résultat indique que la performance entre les deux tâches de compréhension n'est pas corrélée et que par conséquent, les deux tests mesurent des construits différents. La validité divergente de la BCS est donc considérée excellente.

Pour l'établissement de la validité discriminante, une analyse de chi-carré de Pearson bilatéral a été effectuée entre le taux de participants identifiés comme présentant un trouble de compréhension syntaxique à la tâche C-2 de la BCS et le taux de participants identifiés comme présentant un trouble de compréhension syntaxique à la tâche de compréhension orale de phrases du MT-86. Avec le MT-86, 7 participants sur 30 ont été identifiés comme présentant un trouble de compréhension syntaxique, comparativement à 23 participants sur 30 avec la BCS. La BCS a donc été en mesure d'identifier 16 participants de plus que le MT-86 et cette différence est significative, $\chi^2(1, N = 30) = 17{,}07$, $p < 0{,}001$. Ce résultat indique que la BCS est plus sensible que le MT-86 pour identifier les individus présentant un trouble de compréhension syntaxique. Une analyse complémentaire des caractéristiques individuelles des participants quant à leur compréhension syntaxique (atteinte vs préservée) indique également que tous les participants ayant été identifiés comme présentant un trouble de compréhension syntaxique avec le MT-86 ($n = 7$) l'ont également été avec la BCS. Ce résultat indique que la BCS est aussi spécifique que le MT-86 dans l'identification de ces individus, tout en étant plus sensible.

Finalement, pour l'établissement de la fidélité test-retest, une analyse de coefficient de corrélation intra-classe à modèle mixte à deux facteurs de type cohérence absolue a été effectuée, ICC = 0,963, $F(44, 44) = 24{,}947$, $p < 0{,}001$. Ce résultat indique une excellente fidélité test-retest pour la BCS.

Globalement, les résultats de l'analyse de validation suggèrent que la BCS a de bonnes qualités métrologiques.

Phase 3 : normalisation de l'outil

La troisième phase du projet avait pour objectif d'obtenir des données normatives pour la BCS en l'administrant à des participants contrôles.

Procédures de collecte de données

Le projet a été approuvé par le comité d'éthique du centre de recherche CERVO (certificat d'éthique #2015-450). Cent participants adultes, sans trouble cognitif avéré et dont la langue maternelle était le français, ont été recrutés dans deux pays : la Suisse romande ($n = 75$) et le Québec ($n = 25$). Tous les participants présentaient un score ≥ 26/30 au *Montreal Cognitive Assessment* (Nasreddine et al., 2005), indiquant un fonctionnement cognitif normal. Les participants ont rempli une fiche signalétique et aucun d'entre eux n'a rapporté de troubles physiques ou mentaux pouvant interférer avec leur performance cognitive (p. ex : maladie neurologique, trouble visuel ou auditif non corrigé, traumatisme crânio-cérébral etc.). L'échantillon était composé de 54 hommes et 46 femmes, âgés entre 46 et 80 ans (moyenne d'âge = 62,1), avec un niveau d'éducation variant de 7 à 24 ans (scolarité moyenne = 13,2 ans).

Les participants ont été recrutés par des étudiants en logopédie/orthophonie via des annonces publiques et parmi leurs proches. Tous ont été testés individuellement, dans une pièce calme, chez eux ou à l'université, et la BCS a été administrée sans contrainte de temps. Les évaluateurs ont tous été préalablement formés par l'équipe de recherche à la passation standardisée de la BCS.

Résultats

Compte tenu de la provenance diverse de l'échantillon, une première analyse a été effectuée afin de vérifier si ces deux groupes étaient comparables en termes de répartition du sexe des participants, de leur âge et de leur niveau de scolarité. Des analyses de test-t pour échantillons indépendants ont montré une absence de différence entre les groupes, tant pour le sexe des participants, $t(98) = -0{,}118$, $p = 0{,}906$, leur âge, $t(98) = -0{,}038$, $p = 0{,}907$, que pour leur niveau de scolarité, $t(98) = 0{,}858$, $p = 0{,}356$. Ce premier résultat indique que les participants provenant du Québec et de la Suisse avaient des caractéristiques démographiques équivalentes.

Par la suite, les performances aux différentes tâches de la BCS ont été analysées en fonction de la provenance des participants. Des analyses de test-t pour échantillons indépendants ont montré une absence de différence entre les groupes à chaque tâche de la BCS. Le **tableau 2** illustre ces résultats. Par conséquent, les 100 participants ont été regroupés dans un même groupe pour poursuivre les analyses de normalisation.Une fois tous les participants regroupés, une première analyse de corrélation de Pearson a été effectuée entre les résultats aux différentes tâches de la BCS, l'âge et le niveau de scolarité des participants. Une corrélation positive significative a été obtenue entre le niveau de scolarité et le score à aux tâches A-2, $r(98) = 0{,}197$, $p = 0{,}050$, et C-2, $r(98) = 0{,}257$, $p = 0{,}010$. Ce résultat indique qu'il y a une association positive entre le score à ces tâches et le

Tableau 2

Performance aux différentes tâches en fonction de la provenance des participants

Tâche	Moyenne (*ÉT*) Québec	Moyenne (*ÉT*) Suisse	*t*	*dl*	*p*
A-1 : Catégorie grammaticale	19,96 (0,20)	19,85 (0,43)	1,207	98	0,230
A-2 : Jugement de grammaticalité	34,96 (0,94)	34,75 (2,06)	0,500	98	0,619
B : Représentation lexico-argumentale du verbe	9,88 (0,33)	9,83 (0,42)	0,583	98	0,561
C-1 : Pré-test de compréhension lexicale	10,00 (0,00)	9,97 (0,16)	0,819	98	0,415
C-2 : Assignation des rôles thématiques	48,36 (1,19)	47,59 (1,91)	1,901	98	0,060

niveau de scolarité des participants. Une corrélation négative significative a également été obtenue entre le niveau de scolarité des participants et leur âge, $r(98) = -0,357$, $p < 0,001$.

Compte tenu de l'association entre les variables démographiques (âge et scolarité), une analyse de régression linéaire multiple (méthode entrée) a été effectuée pour prédire le score à la tâche C-2 en se basant sur l'âge et le niveau de scolarité. Une équation de régression significative a été obtenue, $F(2,97) = 3,439$, $p = 0,036$, où seul le niveau de scolarité était un prédicteur significatif ($\beta = 0,250$, $p = 0,019$). Comme l'âge n'a encore une fois pas été significativement associé à la performance à la BCS ($\beta = -0,019$, $p = 0,859$), il a été décidé de procéder à une stratification des données de normalisation seulement en fonction du niveau de scolarité. Le seuil de stratification choisi était 12 ans de scolarité complétés, ce qui a permis de former un groupe de participant ayant une faible scolarité (entre 0 et 11 années de scolarité complétées, $n = 29$) et un autre groupe ayant une scolarité élevée (12 années ou plus de scolarité complétées, $n = 71$). Ce seuil a été choisi en s'appuyant sur ce qui a précédemment été effectué dans la littérature scientifique, notamment dans les publications présentant des données normatives d'autres tests orthophoniques (p. ex. Macoir et al., 2017).

Finalement, une dernière analyse de test-t pour échantillons indépendants a été effectuée pour mesurer l'effet du groupe de scolarité (faible scolarité vs scolarité élevée) sur les performances aux tâches de la BCS. Un effet significatif de la scolarité a de nouveau été obtenu pour les tâches A-2, $t(98) = -2,092$, $p = 0,039$, et C-2, $t(98) = -2,896$, $p = 0,005$. Ce résultat indique que la stratification en fonction du niveau de scolarité tel qu'effectuée (seuil à 12 ans) est adéquate et reflète bien la variation de performance selon le niveau de scolarité pour les tâches A-2 et C-2.

Le **tableau 3** présente les données normatives (moyenne et écart-type) en fonction du niveau de scolarité des participants, et ce, pour chaque tâche de la BCS. Le **tableau 3** présente également les points d'alerte (15^{e} percentile), de même que les seuils critiques (5^{e} percentile) pour chaque tâche. Ces percentiles ont été calculés en fonction des scores Z associés à chaque percentile ciblé selon la distribution normale, puis arrondi à l'unité inférieure. À noter que pour certaines tâches, le point d'alerte et le seuil critique correspond au même score, compte tenu de la très faible variabilité des résultats normatifs.

Discussion

L'objectif principal de cet article était de présenter les processus de développement, de validation et de normalisation d'une batterie d'évaluation de la compréhension syntaxique récemment publiée au Québec par le CIUSSS de la Capitale-Nationale – IRDPQ (Caron et al., 2015). La BCS constitue un outil novateur et utile pour analyser de façon objective et exhaustive les différentes étapes de la compréhension de phrases, tout en tenant compte des paramètres psycholinguistiques pouvant influencer la performance des patients. La BCS a été explicitement développée pour répondre au besoin clinique d'un outil d'évaluation spécifique au trouble de compréhension syntaxique rencontré dans les aphasies post-AVC ou neurodégénératives. En effet, contrairement aux outils actuellement disponibles en français, la BCS s'appuie sur un modèle théorique reconnu et qui offre l'avantage de spécifier les différentes étapes de traitement menant à la compréhension des phrases. Chacune des étapes de traitement syntaxique peut donc maintenant être évaluée de manière indépendante, ce qui permet d'identifier l'origine précise des déficits observés chez les personnes ayant un trouble de compréhension syntaxique. En cela, la BCS permet aux orthophonistes d'offrir des interventions plus ciblées et efficaces.

Les résultats de l'étude métrologique de la BCS ont montré que la batterie avait une bonne validité convergente, d'excellentes validités divergente et discriminante, ainsi qu'une excellente fidélité test-retest. Quant à la normalisation, il est intéressant d'avoir obtenu des données sans différence significative entre les populations francophones de la Suisse et du Québec.

Tableau 3

Données normatives (n = 100) pour chaque tâche de la *Batterie d'évaluation de la compréhension syntaxique* (moyenne, écart-type, point d'alerte et seuil critique) en fonction du niveau de scolarité

	Tâche	Score maximal	Moyenne	Écart-type	Point d'alerte (< 15e percentile)	Seuil critique (< 5e percentile)
Faible scolarité (< 11 ans)	A-1 : Catégorie grammaticale	20	19,79	0,49	19	18
	A-2 : Jugement de grammaticalité	36	34,21	2,94	31	29
	B : Représentation lexico-argumentale du verbe	10	9,83	0,38	9	9
	C-1 : Pré-test de compréhension lexicale	10	9,97	0,19	9	9
	C-2 : Assignation des rôles thématiques	50	47,00	2,14	44	43
Scolarité élevée (≥ 12 ans)	A-1 : Catégorie grammaticale	20	19,92	0,32	19	19
	A-2 : Jugement de grammaticalité	36	35,04	1,06	33	33
	B : Représentation lexico-argumentale du verbe	10	9,85	0,40	9	9
	C-1 : Pré-test de compréhension lexicale	10	9,99	0,12	9	9
	C-2 : Assignation des rôles thématiques	50	48,10	1,52	46	45

L'utilisation de l'outil est donc applicable auprès des locuteurs de ces deux variantes de français. L'âge des sujets n'influence pas non plus les résultats, mais la performance est cependant affectée par le niveau d'éducation. Ainsi, les participants avec une scolarité plus élevée ont légèrement mieux performé que ceux détenant moins d'années d'éducation formelle. Le tableau des normes est ainsi stratifié en ce sens (voir le **tableau 3**). Il est intéressant de noter que seules les tâches A-2 et C-2 de la batterie se sont montrées sensibles au niveau d'éducation. En fonction du modèle théorique de compréhension syntaxique sur lequel la BCS est basée, ses tâches correspondent aux étapes d'analyse syntaxique (ou *parsing*) et d'assignation des rôles thématiques (ou *mapping*), lesquelles reflètent des fonctions langagières strictement syntaxiques.

Une des grandes forces de cette étude est le nombre de participants : 100 personnes sans lésion cérébrale ont été évaluées en tant que participants contrôles pour la normalisation, 30 personnes aphasiques l'ont été pour l'évaluation des validités et 9 de ces personnes aphasiques l'ont été à une deuxième reprise pour l'évaluation de la fidélité test-retest. Une limitation qui pourrait être mentionnée dans la présente étude normative est l'utilisation d'une méthode d'échantillonnage accidentel, ce qui aurait pu entrainer un biais de sélection. Une autre limitation concerne le fait que les étapes de validation ont été réalisées auprès de seulement 30 personnes aphasiques d'origine québécoise. La normalisation, quant à elle, a été réalisée auprès des deux populations (franco-suisse et franco-québécoise). Toutefois, étant donné que les normes n'ont pas montré de différence entre les deux provenances géographiques, il est permis de croire qu'il en aurait été de même pour les participants recrutés pour la validation. Enfin, parmi les limitations, il n'a pas été possible de trouver, parmi les tests existants en français, des tâches comparables à celles A-1, A-2 et B de la BCS. Seules les tâches C-1 et C-2 ont pu être comparées avec des tâches précédemment publiées dans le but d'établir la validité convergente de la BCS. Or, ce qui peut apparaître comme une limitation peut également être vu comme une force, puisqu'il s'agit d'un nouvel outil comportant des tâches inexistantes ailleurs. Enfin, il est intéressant de mentionner que les améliorations apportées grâce à l'étape de validation de contenu permettent d'assurer que l'outil est fonctionnel et facile d'utilisation, que le temps de passation et de cotation est adéquat et que les images et les feuilles de cotation ne sont pas ambiguës.

Finalement, même si la BCS a été conçue en s'appuyant sur un modèle conceptuel de compréhension syntaxique et qu'il permet théoriquement d'étudier les patrons d'erreurs précis en regard de ce modèle, le présent article n'aborde pas cette question. Cette décision a été prise, puisque les objectifs de l'article étaient 1) de présenter le processus de développement, de validation et de normalisation de l'outil et 2) de présenter les données normatives. Il est néanmoins à noter qu'un second article, actuellement en préparation, visera à analyser les patrons d'erreurs des participants en les mettant en relations avec les modèles théoriques actuels de compréhension syntaxique (p. ex. analyse syntaxique ou *parsing* vs. assignation des rôles thématiques ou *mapping*).

Conclusion

En somme, le développement, la validation et la normalisation d'un nouvel outil d'évaluation de la compréhension syntaxique, comme la BCS, constituent des avancements majeurs dans la pratique clinique orthophonique. En effet, pouvoir mieux cibler l'origine du déficit de compréhension de phrases permet conséquemment au clinicien de choisir avec plus de précision les activités d'intervention et optimise ainsi le rétablissement. Il serait d'ailleurs intéressant dans le futur de développer des méthodes d'intervention spécifiques liées aux étapes de la BCS et d'en mesurer les effets.

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Spatial Processing Disorder in Children With Cleft Palate

Le trouble du traitement auditif relié à la spatialité chez les enfants ayant une fissure palatine

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Abstract

Spatial Processing Disorder is manifested as difficulty understanding speech in a noisy environment despite normal standard audiometric results. Rates of Spatial Processing Disorder are significantly higher in children who have a history of otitis media in early childhood, and the prevalence and duration of otitis media in children with cleft palate are significantly higher than the general population. Therefore, this study aimed to determine the prevalence of Spatial Processing Disorder in this vulnerable population. Children with cleft palate aged between 6 and 16 were recruited from a cleft palate clinic. Those with normal audiograms and absence of ear disease, communication disorders, or intellectual disability were included in the study. Eight (40%) of the 20 children who met inclusion criteria were found to have Spatial Processing Disorder using the Listening in Spatialized Noise-Sentences diagnostic standard. Four additional patients were found to have signal-to-noise ratio losses greater than 2 dB from the mean, representing substantial loss in speech intelligibility. Three children underwent remediation using the Listening in Spatialized Noise & Learning program; all saw substantial benefit. Spatial Processing Disorder may be of detriment particularly in school-aged children and is highly prevalent in those with cleft palate. As therapy for this disorder has recently been developed, prompt identification and intervention may improve the learning experience of affected children.

Abrégé

Le trouble du traitement auditif relié à la spatialité se manifeste par une difficulté à comprendre la parole dans un environnement bruyant, et ce, malgré des résultats normaux aux tests audiométriques standards. La proportion d'enfants ayant un trouble du traitement auditif relié à la spatialité est significativement plus élevée chez ceux ayant un historique d'otites moyennes survenues durant les premières années de vie. Ajoutons que la prévalence et la durée des otites moyennes sont significativement plus élevées chez les enfants ayant une fissure palatine que chez les enfants provenant de la population générale. La présente étude visait donc à déterminer la prévalence d'enfants ayant un trouble du traitement auditif relié à la spatialité au sein des enfants ayant une fissure palatine. Des enfants ayant une fissure palatine âgés entre 6 et 16 ans ont été recrutés dans une clinique de fissure palatine. Les enfants ayant des audiogrammes normaux, en l'absence d'un problème otologique, d'un trouble de communication ou d'une déficience intellectuelle, ont été inclus dans l'étude. Huit (40%) des 20 enfants qui respectaient les critères d'inclusion avaient un trouble du traitement auditif relié à la spatialité en utilisant le test *Listening in Spatialized Noise-Sentences*. Quatre autres enfants avaient une perte du ratio signal/bruit supérieure à 2 dB par rapport à la moyenne, ce qui représente une perte importante de l'intelligibilité de la parole. Une intervention utilisant le programme *Listening in Spatialized Noise & Learning* a été offerte à trois des enfants; tous en ont grandement bénéficié. La présence d'un trouble du traitement auditif relié à la spatialité peut porter préjudice aux enfants en étant atteints, et ce, particulièrement à l'âge scolaire. Ajoutons que sa prévalence est élevée chez les enfants ayant une fissure palatine. Étant donné qu'un programme d'intervention a récemment été développé pour ce trouble, une identification et une intervention rapides peuvent améliorer l'expérience d'apprentissage des enfants ayant un trouble du traitement auditif relié à la spatialité.

Spatial Processing Disorder (SPD) can be considered a type of Central Auditory Processing Disorder in which patients with normal standard audiometric test results have listening difficulties (Moore, Rosen, Bamiou, Campbell, & Sirimanna, 2013). Patients with SPD have difficulty attending to streams of speech in noisy environments due to inability to select sounds from a single direction when there is extensive competing background noise (Schneider, 2011). SPD can be debilitating for many patients, but is disproportionately significant in children given their attendance in often-noisy school environments (Mealings, Demuth, Buchholz, & Dillon, 2015).

Recently, a test designed to identify and diagnose SPD has been developed at the National Acoustics Laboratory in Australia (Cameron & Dillon, 2008). This test, the Listening in Spatialized Noise-Sentences (LiSN-S) test, is a virtual, computer-based assessment tool that measures the ability of patients to use spatial cues to differentiate a target talker from competing talkers. Patients with inability to rely on inter-aural cues will predictably perform poorly on spatially separated conditions compared to their counterparts. The effects of non-auditory factors, such as cognitive abilities, attention, and language, are minimized by making the relevant score a "difference" measure between scores on subtests with different acoustic characteristics.

SPD has not been extensively studied, especially in the pediatric population. Preliminary evidence has shown that rates of SPD are significantly higher in children who have a chronic history of otitis media in early childhood (Dillon, Cameron, Glyde, Wilson, & Tomlin, 2012). This suggests that even transient auditory deprivation over time may contribute to the development of SPD due to under stimulation of the central auditory nervous system. In other words, the development of a child's spatial auditory processing abilities could be disrupted by fluctuating hearing levels during critical early developmental periods. Indeed, the prevalence of SPD was found to be significantly higher in Australian Indigenous children, a group with substantially higher rates of otitis media (Cameron, Dillon, Glyde, Kanthan, & Kania, 2014).

Otitis media is common among children with cleft palate. The likelihood of having otitis media by 2 years of age is 97% for children with cleft palate, compared to 60% for the general population (Kuo, Lien, Chu, & Shiao, 2013). Although the incidence of otitis media in this population decreases over time, many children can have chronic Eustachian tube dysfunction and the prevalence of SPD has yet to be examined in children with cleft palate.

The developers of LiSN-S have also created a virtual rehabilitation program designed to improve children's spatial processing abilities and treat SPD. Listening in Spatialized Noise & Learning (LiSN & Learn) employs a series of computer games trained over a period of time, with results showing nearly a full standard deviation of improvement in terms of spatial advantage by the completion of the intervention (Cameron et al., 2014).

This study aimed to (a) assess the prevalence of SPD in children with cleft palate, (b) describe their spatial processing ability, and (c) examine the possibility of SPD rehabilitation using an established remediation program.

Method

This study was approved by the local Research Ethics Board (Izaac Walton Killam Health Centre Protocol # 1020364).

Participants

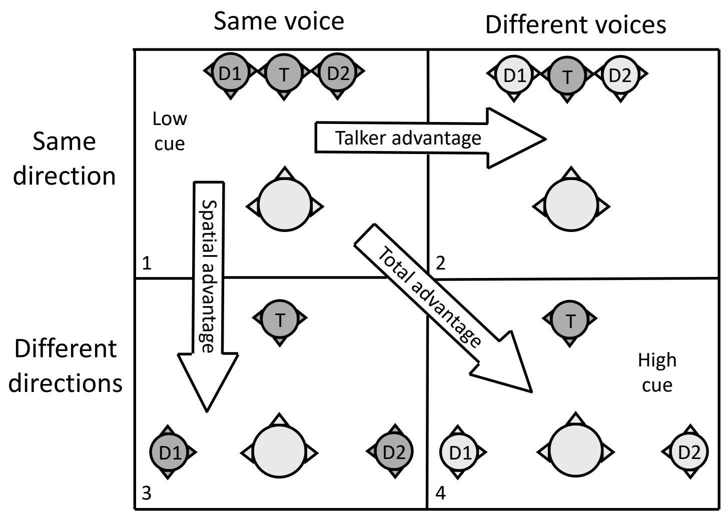
Children with cleft palate (with or without cleft lip) between the ages of 6 and 16 years were recruited from the cleft palate clinic at the Izaac Walton Killam Health Centre in Halifax, Nova Scotia. The age limits were chosen as children under 6 years cannot be reliably tested for SPD using the LiSN-S program (Brown, Cameron, Martin, Watson, & Dillon, 2010). Other inclusion criteria included normal hearing thresholds from 500 Hz to 4000 Hz in both ears and fluency in English. Exclusion criteria included (a) children with intellectual disability, autism spectrum disorder, or with documented, un-treated attention deficit or hyperactivity disorders and (b) abnormal otoscopic examination with the presence of external or middle ear disease.

Medical records of participants were reviewed to obtain demographics, frequency of otitis media diagnoses, and past history of tympanostomy tubes.

Listening in Spatialized Noise-Sentences Test

Participants underwent the LiSN-S test as previously described (Cameron et al., 2009). Briefly, the signal-to-noise ratio (SNR) required to identify 50% of the words in target sentences is established in four conditions that vary in terms of the virtual location of the noise source and the vocal quality of the speaker (**Figure 1**). All sounds are played through a standardized headphone system that stimulates spatialized sound. The target is always presented at 0° azimuth, while competing talkers are presented at 0° (no spatial separation) or at 90° spatial separation. The competing talkers can be the same voice as the target (i.e., no voice cues) or they can be voices different from the target. Using a combination of these parameters, four conditions are created:

Figure 1



The four conditions of the Listening in Spatialized Noise-Sentences test, numbered according to the description in the text in the Method section. Adapted from Cameron, Dillon, Glyde, Kanthan, and Kania (2014). D1 and D2 represent the competing (or distracting) talkers, and T represents the target talker. When D1, D2, and T are the same colour, the voice quality is identical. When D1, D2, and T are different colours, the voice quality is different.

1. Low-cue condition (competition is the same voice at 0° azimuth).
2. Different voices condition (competition is a different voice at 0° azimuth).
3. Spatial separation only condition (competition is the same voice at 90° azimuth).
4. High-cue condition (i.e., competition is a different voice at 90° azimuth).

Subtracting the score for one condition from the score for another results in difference scores. For the purposes of this study, the key difference score is the difference between Condition 3 and Condition 1. This difference score is defined as the *spatial advantage* since the only difference between Condition 1 and Condition 3 is the addition of spatial cues in Condition 3. Less important is the *talker advantage* score, which is defined as the difference between Condition 1 (same voice competition at 0° azimuth) and Condition 2 (different voice competition at 0° azimuth).

Spatial processing disorder was diagnosed using the LiSN-S test if the spatial advantage score was equal to or greater than 2 standard deviations (*SD*) lower than the North American mean (Cameron et al., 2009).

In addition to analyzing data in terms of deviance from the mean, the LiSN-S program also provides the clinician with quantity of SNR loss, if any. For example, if a spatial advantage of 9.0 dB was expected, and the participant's spatial advantage was 6.5 dB, then the participant's SNR loss would be 2.5 dB. Clinically meaningful decrements in SNR advantage were examined, even if the 2 *SD* criterion was not met. A clinically meaningful decrement was defined as an SNR advantage 2 dB below the age-specific mean, since this corresponds to a noticeable difference in a child's ability to listen in a noisy environment (Killion, 2004).

Listening in Spatialized Noise & Learn Training

Children diagnosed with poor performance during the early study period were offered a chance to complete the LiSN & Learn training outside of the hospital environment, either at home or at school. The training protocol has previously been described (i.e., Cameron & Dillon, 2011; Cameron et al., 2014; Cameron, Glyde, & Dillon, 2012). Briefly, children play two auditory training games per day for 5 days each week, for 50 total training sessions or 100 total games (total of 15–20 minutes per day). In all games, the child identifies a word from a target sentence while competing sentences are presented at either 0° azimuth or ± 90° azimuth. Correct answers result in progressively more difficult games in the form of decreasing target voice levels. For incorrect answers, the target voice level is raised.

Data Analysis

Simple descriptive statistics were utilized for quantifying patient characteristics and determining variance from population means for spatial and talker advantages. Two-tailed Student's *t* tests were used for statistical analysis of continuous variables. Correlations were determined using the Pearson correlation coefficient.

Results

Participants

Twenty patients (10 girls) during the study period (October 2015 and October 2016) met inclusion criteria. The median age was 9 years, and all patients had previously repaired cleft palate, with or without cleft lip. The median age at time of cleft palate repair was 11.0 months.

Listening in Spatialized Noise-Sentences Test

Low-cue and high-cue speech reception threshold (SRT) measurements, talker advantage measurements,

Table 1

Raw Listening in Spatialized Noise-Sentences Data for All Patients[1]

Patient	Low-cue SRT	High-cue SRT	Talker advantage	Spatial advantage
01	-0.2	-10.2	2.7	8.6
02	-0.8	-14.6	8.1	13.4
03	1.1	-9.1	2.5	8.3
04	5.1	2.9	2.7	-3.1
05	-2.1	-10.9	3.6	6.1
06	0.6	-14.2	5.6	13.6
07	0.4	-11.1	6.6	8.0
08	1.3	-9.2	4.2	8.1
09	0.0	-6.8	2.9	4.7
10	1.6	-5.2	1.7	4.2
11	2.3	-6.8	3.0	7.8
12	1.9	-8.5	2.5	4.1
13	0.9	-7.5	5.5	6.4
14	7.6	3.6	5.0	4.6
15	0.2	-10.4	1.8	6.8
16	-0.2	-6.8	3.1	5.8
17	0.9	-9.5	4.2	9.6
18	2.4	-7.0	1.1	8.7
19	1.6	-6.2	3.4	6.5
20	-0.1	-8.1	6.1	8.0

Note. [1]All measurements in decibels (dB). SRT = Speech Reception Threshold.

and spatial advantage measurements for all participants are listed in **Table 1**. There were no statistically significant correlations between patient age and low-cue (same voice) SRT, $r(19) = -.50$, $p > .05$; high-cue (different voice) SRT, $r(19) = -.56$, $p > .05$; talker advantage, $r(19) = .45$, $p > .05$; or spatial advantage, $r(19) = .53$, $p > .05$. Similarly, there were no significant gender-related correlations, $r(19) = .33, .42, -.18$, and $-.37$, respectively, all $p > .05$. There was a strong, significant positive correlation between low-cue SRT and high-cue SRT, $r(19) = .85$, $p < .05$, but not between low-cue SRT and talker advantage or spatial advantage. However, there was a strong, significant negative correlation between high-cue SRT and spatial advantage, $r(19) = -.80$, $p < .05$. The frequency of myringotomy and tympanostomy tube placement did not correlate with any LiSN-S measurements.

Spatial Processing Disorder Diagnosis

Eight patients (40%) were diagnosed with SPD (**Figure 2**). Raw data is available in the Appendix. The median age was 8 years 5 months, with a slight (non-significant) female predominance (62.5%). The characteristics of patients diagnosed with SPD are found in **Table 2**. There was no difference in mean age at time of cleft palate repair between those diagnosed with SPD and those who were not (20.1 vs. 26.0 months, $p > .05$). There was no difference in the mean number of tympanostomy tube procedures required between the two groups (2.2 in those without SPD, 2.9 for those with SPD, $p > .05$).

There were no correlations between variance from the spatial advantage mean and talker advantage scores or talker advantage variance from the mean. The majority of patients were within 2 *SD* of the population mean for their talker advantage measurement (91.7%).

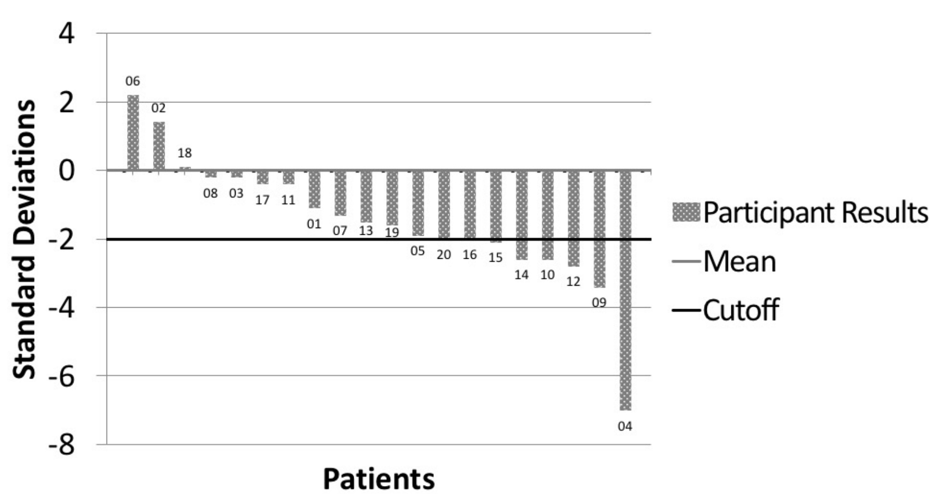
Four patients who were not diagnosed with SPD were found to have a meaningful (> 2 dB) increase in SNR thresholds, resulting in a total of 12 patients (60%) with significant difficulties understanding speech in noise (see **Figure 3**).

Listening in Spatialized Noise & Learn Training

Two children diagnosed with SPD and one child with meaningful increase in SNR thresholds underwent spatial processing remediation with the LiSN & Learn software. Not all eligible participants were offered training due to lack of availability of the remediation software and some did not have the ability to use the program (i.e., lack of appropriate hardware).

The results of remediation training are presented in **Table 3**. All three children had improvement in their

Figure 2



Variance of spatial advantage from the standardized North American population mean for all participants. The cutoff of two standard deviations from the mean was considered significant for the diagnosis of Spatial Processing Disorder. Values above and below the data bars represent the patient ID; for example, the bar to the furthest right is Patient 04. Positive scores represent patients scoring better than the population mean, while negative are worse.

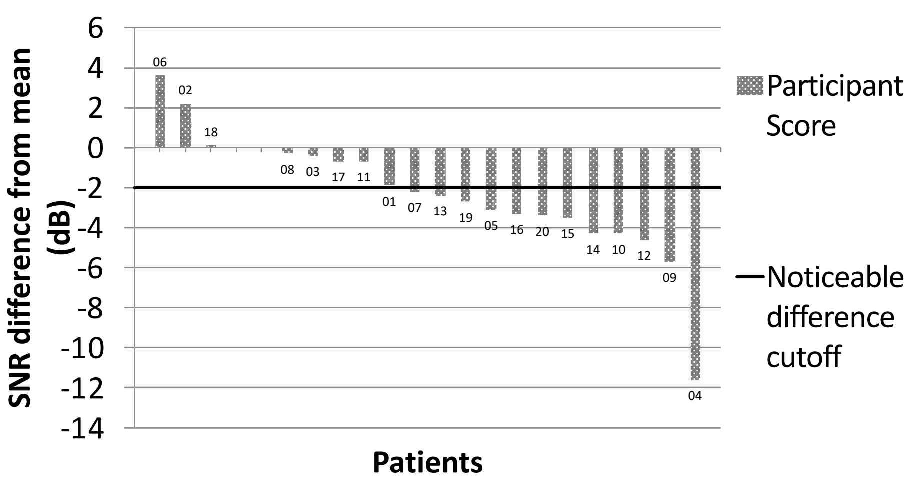
Table 2

Characteristics of Patients Diagnosed With Spatial Processing Disorder

Patient	Age (months)	Diagnosis	Repair	Ear history	Other
04	81	Cleft lip & palate	Yes	M & T x 2	
05	145	Cleft palate	Yes	M & T x 5	
10	82	Bilateral cleft lip & palate	Yes	M & T x 3	
12	91	Cleft lip & palate	Yes	M & T x 3	Loss of alveolar bone graft secondary to trauma
14	97	Unilateral cleft lip & bilateral palate	Yes	OME, COM, No M & T	
15	144	Cleft palate	Yes	M & T x 1	Congenital unilateral blindness
16	104	Submucosal cleft palate	Yes	No M & T	
20	113	Cleft palate	Yes	M & T x 6	

Note. M & T = myringotomy and tympanostomy tubes; OME = otitis media with effusion; COM = chronic otitis media.

Figure 3



Signal-to-Noise ratio differences in the spatial advantage measurement of all participants. The noticeable difference cutoff was defined as two decibels. Values above and below the data bars represent the patient ID; for example, the bar to the furthest right is Patient 04. Positive scores represent patients scoring better than the population mean, while negative are worse.

Table 3

Spatial Advantage Measurements Before and After Remediation With the Listening in Spatialized Noise & Learning Program

Patient	Spatial advantage - pre (dB)	Spatial advantage - post (dB)	Spatial advantage - improvement (dB)	Spatial advantage variance from mean - pre (*SD*)	Spatial advantage variance from mean - post (*SD*)	Spatial advantage variance from mean -improvement (*SD*)
04	-3.10	9.10	12.20	-7.00	0.10	7.10
05	6.10	10.00	3.90	-1.90	0.20	1.70
10	4.20	5.30	1.10	-2.60	-1.70	0.90

Note. dB = Decibels.

spatial advantage and were considered to be within normal range following the intervention.

Discussion

Spatial processing disorder is an under-recognized clinical entity that disproportionately affects children with a history of otitis media. The prevalence of SPD was found to be higher in our study population (40%) than in Indigenous Australian children (7%) who are also thought to be at higher risk for otitis media (Cameron et al., 2014). However, despite having potentially the highest rates of middle ear disease among ethnic groups, rates of otitis media may be higher in the cleft palate population than the Indigenous Australian children. Prevalence of middle ear disease in the Indigenous population has been reported to be as high as 62% (Thorne, 2004), yet rates of otitis media reach nearly 100% in cleft palate patients (Dhillon, 1988). This may explain why the prevalence of SPD was higher in our study. There was a slight female predominance in our study that was not statistically significant. This is likely due to the small population size, as boys are often quoted as having a higher rate of cleft lip and palate than girls.

In addition to the high prevalence of SPD in our patient population, nearly all patients had SNR thresholds that were higher than age-appropriate means. An SNR of only 1 dB above the mean may result in a speech intelligibility loss of up to 17% in noisy environments (Cameron & Dillon, 2007). While SPD may be debilitating for children in learning environments, the loss in speech intelligibility may also be limiting for these children even though they do not formally meet diagnostic criteria for SPD. On the severe end of the spectrum, the patient with 12 dB increase above the mean could be expected to have almost zero sentence recognition in noisy environments.

The lack of correlation between talker advantage and spatial advantage suggests that the scores do not simply reflect higher order language, learning, and communication skills, which would be expected to affect both to a similar degree, but rather that they reflect specific auditory processes. High-cue SRT and spatial advantage measurements had a strong negative correlation, indicating that children with strong spatial listening skills can also take advantage of talker voice differences to improve hearing in noise.

We have also demonstrated the efficacy of LiSN & Learn remediation training in this population, supporting its use and possible future expansion to other patients with SPD. All children showed spatial advantage scores that improved to the normal range following intervention. This is in keeping with the findings of Cameron et al. (2014), in which Indigenous Australian children saw improvements of 1 *SD* on average. One of the children in our study had substantial changes in his or her variance from the population mean, while the other two showed smaller improvements similar to those found in Cameron et al. (2014). The latter participant (Patient 04) was a considerable outlier in terms of both raw spatial advantage score (**Table 1**) and SRT improvement (**Table 3**). Given the small population size in this study, this outlier may have affected correlation calculations. However, many of the LiSN-S correlations are in keeping with expected results and therefore it is unlikely that the outlier caused any substantial statistical aberrations. An earlier randomized, blinded study (i.e., Cameron et al., 2012) reported average SRT reductions of 10 dB. Despite all of the patients in our study having similar *SD* changes as previously reported, only the outlier patient reached the level of dB improvement seen in the trial of Cameron et al.

To our knowledge, the current study is the first to investigate the prevalence of SPD in patients with cleft palate. Despite the small sample size, this study represents all patients from a large geographical area in the Maritime provinces of Canada. Moreover, at the time of testing, all patients had normal otoscopic examinations, normal audiometric testing, and normal tympanometry, so results cannot be attributed to transient differences in middle ear function. Another limitation is that not all participants underwent remediation training due to logistical reasons (i.e., lack of access to the proper equipment to run the LiSN & Learn program), thereby limiting our rehabilitation population size to a small cohort. However, our results are consistent with previous findings that there is improved spatial processing after training.

Future studies should investigate the prevalence of SPD in a larger population of patients with cleft palate, as well as evaluate the effectiveness of the LiSN & Learn program in a larger sample from this population. This study is at low risk for Type II error as the null hypothesis of there being no difference in rates of SPD for patients with cleft palate was rejected. We have limited the Type I error by choosing an alpha of .05 *a priori,* despite the rarity of SPD. Of course, increasing the sample size by a large degree in the future would allow for a stricter *a priori* setting of alpha. It would also be helpful to determine whether training-related improvements in spatial advantage, as found using the LiSN-S, correspond to improvements in functioning in school or in other real world noisy environments.

In summary, nearly half of cleft palate patients in our study population, despite medical and surgical intervention, were found to have spatial processing difficulties and therefore could have significant hearing issues when in a noisy environment. Given the improvements demonstrated by the use of the LiSN & Learn training program, albeit in a limited sample, these difficulties may be amenable to remediation. This is the first study to investigate the prevalence of SPD in patients with cleft palate and the first to demonstrate the potential remediation of SPD in this group.

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Appendix

Raw Variance Data From Listening in Spatialized Noise-Sentences Testing

Patient ID	Low-cue SRT		High-cue SRT		Talker advantage		Spatial advantage		
	Variance from mean LCSRT score	Variance from mean in *SD*	Variance from mean HCSRT score	Variance from mean in *SD*	Variance from mean TA score	Variance from mean in *SD*	Variance from mean SA score	Variance from cutoff score	Variance from mean in *SD*
01	0.9	-0.9	2.2	-1.1	-4.2	-1.9	-1.9	1.4	-1.1
02	0.7	-0.8	-1.3	0.6	0.4	0.1	2.2	5.6	1.4
03	1.0	-1.0	0.6	-0.3	-2.0	-0.9	-0.4	2.9	-0.2
04	4.8	-5.0	12.3	-5.9	-1.5	-0.7	-11.6	-8.2	-7.0
05	-1.9	1.9	-0.3	0.2	-1.6	-0.7	-3.1	0.2	-1.9
06	1.3	-1.4	-2.5	1.2	-0.6	-0.3	3.6	6.9	2.2
07	1.2	-1.3	0.8	-0.4	0.2	0.1	-2.2	1.2	-1.3
08	1.0	-1.0	0.1	-0.1	0.1	0	-0.3	3.0	-0.2
09	1.0	-1.0	5.4	-2.6	-3.8	-1.7	-5.7	-2.3	-3.4
10	1.3	-1.3	4.2	-2.0	-2.5	-1.1	-4.3	-0.9	-2.6
11	2.0	-2.1	2.6	-1.3	-1.2	-0.6	-0.7	2.7	-0.4
12	1.8	-1.9	1.3	-0.6	-1.9	-1.0	-4.6	-1.3	-2.8
13	0.9	-0.9	2.5	-1.2	1.2	0.4	-2.4	0.9	-1.5
14	7.6	-7.9	13.7	-6.6	0.2	0.1	-4.3	-1.0	-2.6
15	1.2	-1.2	1.7	-0.8	-4.8	-2.2	-3.5	-0.2	-2.1
16	0	0	3.6	-1.7	-2.0	-0.9	-3.3	0	-2.0
17	1.8	-1.9	2.5	-1.2	-2.4	-1.1	-0.7	2.7	-0.4
18	2.2	-2.3	2.6	-1.3	-3.3	-1.5	0.1	3.4	0.1
19	1.8	-1.9	4.3	-2.1	-1.8	-0.8	-2.7	0.6	-1.6
20	0.3	-0.3	2.8	-1.3	0.6	0.3	-3.4	0	-2.0

Note. SRT = Speech Reception Threshold; LCSRT = Low Cue Speech Reception Threshold; HCSRT = High Cue Speech Reception Threshold; TA = Talker Advantage; SA = Spatial Advantage.

Investigation of the Psychometric Properties of the Milestones en français du Québec, a New Language Screener for French-Speaking Children Between 12 and 71 Months

Investigations des propriétés psychométriques du questionnaire *Milestones en français du Québec*, un nouvel outil de dépistage des difficultés langagières pouvant être utilisé avec des enfants francophones âgés entre 12 et 71 mois

KEYWORDS

LANGUAGE DEVELOPMENT

DEVELOPMENTAL LANGUAGE DISORDER

SCREENER

PARENT QUESTIONNAIRE

FRENCH

PRESCHOOLERS

VALIDATION

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Abstract

Very few validated screening tools exist for Developmental Language Disorder in Canadian French. This article presents the steps of the development of a new screening questionnaire designed for children ranging in age from 12 to 71 months. Phase A describes how the questionnaire Milestones en français du Québec (MilBec) was elaborated based on the 26-item Dutch questionnaire by Luinge, Post, Wit, and Goorhuis-Brouwer (2006). The elaboration involved translation, adaptation and addition of items, as well as a cross-sectional pilot study with 26 participants aged 1 year (n = 9), 3 years (n = 8), and 5 years (n = 9), leading to a revised version with 39 items. Phase B presents a cross-sectional study in which parents of 85 monolingual French-speaking children aged 12 to 71 months (17 participants per 12-month age group) filled out the MilBec. The correlation between MilBec score and age was extremely high for 12- to 39-month-old children (n = 42; r = .92, p < .001) and high for 40- to 71-month-old children (n = 43; r = .60, p < .001). High scores were observed from age 3 years and a ceiling effect was present at age 5. The MilBec's internal consistency was very high (Cronbach's alpha = .967). Further exploration of the MilBec's psychometric properties, notably its screening accuracy using larger groups of children that are more representative of the general population including varied socioeconomic status and bilingual children, is warranted.

Abrégé

Très peu d'outils normés en français canadien sont disponibles pour le dépistage du trouble développemental du langage. Le présent article décrit la démarche ayant été utilisée pour développer un nouveau questionnaire de dépistage pouvant être utilisé avec des enfants âgés entre 12 et 71 mois. La phase A de l'article rapporte comment le questionnaire néerlandais composé de 26 items et créé par Luinge, Post, Wit et Goorhuis-Brouwer (2006) a été utilisé pour élaborer le questionnaire *Milestones en français du Québec* (MilBec). Le processus d'élaboration a consisté en la traduction, l'adaptation et l'ajout d'items, ainsi qu'en la réalisation d'une étude pilote transversale auprès de 26 participants âgés de 1 an (*n* = 9), 3 ans (*n* = 8) et 5 ans (*n* = 9). Ce processus a mené à la création d'une version révisée comprenant 39 items. La phase B de l'article présente les résultats d'une étude transversale dans laquelle il a été demandé à 85 parents d'enfants francophones unilingues âgés entre 12 et 71 mois (17 participants par tranche d'âge de 12 mois) de compléter le MilBec. La corrélation entre l'âge des enfants et leur score au MilBec était extrêmement élevée pour la tranche d'âge 12–39 mois ($n = 42$; $r = 0{,}92$, $p < 0{,}001$) et élevée pour la tranche d'âge 40–71 mois ($n = 43$; $r = 0{,}60$, $p < 0{,}001$). Des scores élevés au MilBec ont été observés à partir de l'âge de 3 ans et un effet de plafond était présent à l'âge de 5 ans. La cohérence interne du MilBec était très élevée (alpha de Cronbach = 0,967). Ces résultats justifient une exploration plus approfondie des propriétés psychométriques du MilBec, notamment une exploration de la précision du dépistage à l'aide de groupes d'enfants plus nombreux et plus représentatifs de la population générale (p. ex. qui incluent des enfants bilingues et de statuts socioéconomiques différents).

This article presents the first steps undertaken to develop a parent questionnaire to screen for Developmental Language Disorder (DLD) in French-speaking children from the province of Québec, Canada. DLD occurs when the child's language skills are below age expectation and these lower skills impact on the child's life, either in his or her daily communications with others or school achievement. Furthermore, these difficulties are associated with a poor prognosis in the absence of appropriate intervention. If the child has another diagnosis known to cause lowered language skills (e.g., deafness, intellectual disabilities, autism spectrum disorder or other various syndromes), the child is said to have a *language disorder associated with X* (Bishop, Snowling, Thompson, Greenhalgh, & CATALISE-2 consortium, 2017). If the child has no such diagnosis, the child is said to have a *Developmental Language Disorder* even in the presence of co-occurring disorders (e.g., attention deficits, emotional disorders) or risk factors (e.g., parents with low level of education; Bishop et al., 2017). Other children may also have *Speech, Language and Communication Needs* when their language difficulties are related to a physical condition or a limited knowledge of the language of schooling (Bishop et al., 2017). The use of the term DLD instead of other commonly used terms like *Specific Language Impairment* and *Primary Language Impairment* has been strongly advocated to promote consistency; this article follows that recommendation. The prevalence rate of DLD (termed Specific Language Impairment in the cited studies) has been estimated to be around 7.4% for kindergarten children in the United States and found to be influenced by gender and parental education (Tomblin et al., 1997). It was reported to be as high as 9%–14% in a preliminary prevalence study conducted with 5-year-old francophone children in the province of Québec (Elin Thordardottir, 2010; Elin Thordardottir et al., 2003–2008). It is thus a commonly occurring disorder, making early identification crucial.

The goal of a language screening tool is to separate children into two groups: those receiving a score above the cutoff are considered "not at risk" of DLD, whereas those who fail the screening by receiving a score below the cutoff are considered "at risk." The group of children failing a language screening then needs to be further tested by a Speech-Language Pathologist (S-LP) to rule in or rule out the presence of DLD. A multidisciplinary evaluation might be required to assess whether other developmental domains are involved. The new parent questionnaire introduced in this article was developed to screen preschool children for DLD.

There is currently no screening tool validated for French-speaking preschoolers living in the province of Québec, Canada. This reflects the generalized lack of validated tools for this population (Garcia, Paradis, Sénécal, & Laroche, 2006; Gaul Bouchard, Fitzpatrick, & Olds, 2009; Monetta et al., 2016), a lack particularly important for children under the age of 4 years. Indeed, only two tools have been reported as having documented validity for French-speaking children in this age range in the province of Québec (Monetta et al., 2016), namely the parent-questionnaire *Les Inventaires MacArthur-Bates du développement de la communication* (Trudeau, Frank, & Poulin-Dubois, 1999) and *Échelle de vocabulaire en images Peabody* (Dunn, Thériault-Whalen, & Dunn, 1993), neither of which is a screening tool. In the province of Québec, S-LPs may consider using one of several available European screening tools in French, but these tools must also be assessed to determine if their content and norms are appropriate for French Quebeckers. Indeed, Frisk et al. (2009) found that the cutoff scores of American screening tools must sometimes be modified to maximize their sensitivity and specificity when used with Canadian children. These authors suggested that these modifications might be required because of demographic and educational differences between the two countries. It is thus reasonable to assume that similar modifications might be required for European screening tools to be used in the province of Québec.

The purpose of this study was to address the need for a French language screening tool validated in the province of Québec. Phase A presents the selection and adaptation procedures of a parent questionnaire to screen for DLD, as well as a cross-sectional study aimed at identifying elements from a pilot version of the adapted questionnaire that could be improved. Phase A concludes with the presentation of the final version of the questionnaire, the Milestones en français du Québec (MilBec; Paul & Elin Thordardottir, 2010). Phase B presents a cross-sectional study aimed at collecting preliminary data on the MilBec's psychometric properties from a small homogeneous group of monolingual French-speaking children.

Phase A: Adaptation of the Parent Questionnaire Into French

Method

Selection of the tool to adapt. The first step was to determine whether an adaptation of an already existing screening tool was possible or if a new one should be created. To be considered a good candidate for adaptation, the language screening tool should have the following characteristics:

1. The content targets several language domains since DLD is characterized by heterogeneity in its manifestation and can affect many domains of language (Bishop et al., 2017; Leonard, 2014).

2. The administration and scoring procedures should not require specialized training to maximize its potential use by preschool teachers and school-based personnel as well as health care professionals involved in monitoring children's development, whose implication in language screening has been recommended since the 1980s (King & Glascoe, 2003; Tervo & Balaton, 1980).

3. The administration and scoring time should be brief, ideally less than 10 minutes, to be adequate for large-scale screening. It must be noted that in order to be appropriate for large-scale screening, the screening tool also needs to be appropriate for the general population in regards, notably, to ethnicity and socioeconomic status (SES).

4. The targeted age range should cover a reasonably large one, bearing in mind that the need for a screening tool is the highest for children under 48 months. Indeed, as Monetta et al. (2016) reported, for children from the age of 4 years there are more French assessment tools validated in the province of Québec. The large age range and the short administration time were also considered important to reduce the cost and difficulty related to managing multiple versions of the tool.

5. If more than one screening tool was found, the screening tool with the best documented psychometric properties should be favoured. Parent questionnaires were considered particularly good candidates, as they usually entail no specialized training and have a reduced administration and scoring time. Furthermore, several studies support the validity of such measures in many languages for the purpose of documenting language development, thus allowing the identification of children whose development is slower than expected (e.g., Boudreault, Cabirol, Trudeau, Poulin-Dubois, & Sutton, 2007; Elin Thordardottir & Ellis Weismer, 1996; Klee et al., 1998; Marchman & Martinez-Sussmann, 2002).

A review of available European French screening tools was performed to compare their characteristics to the list of criteria. Several screening tools for young children were found (a Quebec French adaptation of the MacArthur-Bates Communicative Development Inventories by Fenson et al., 1993; *Dépistage et Prévention du Langage à 3 ans* introduced in Coquet & Maetz, 1997; *Langage et comportement-3 ans ½* by Chevrie-Muller, Goujard, Simon, & Approche neuropsychologique des apprentissages chez l'enfant, 1994; *Épreuves de repérage des troubles du langage utilisables lors du bilan médical de l'enfant de 4 ans* by Roy, Maeder, & Alla, 1999; the *Batterie rapide d'évaluation des fonctions cognitives* introduced in Billard et al., 2001; *Bilan de santé évaluation du développement pour la scolarité 5-6 ans* introduced in Zorman & Jacquier-Roux, 2002; *Protocole d'Évaluation Rapide* by Ferrand, 2000), but these tools did not fulfill one or more of the targeted characteristics (see Vallée & Dellatolas, 2005, and Société Française de Pédiatrie, 2007, for more details on these tools). The criterion most often unfulfilled was the age range, with many tools targeting an age range of 3 to 9 months only.

Given that a suitable screening tool in French was not found, the second step was a search for screening tools from other languages that could be found in journal articles—and for which a translation of the items was available in French or English. A sixth criterion applied to those potential candidates: the items should mostly target general language milestones (e.g., the age at which babbling starts, the period when two-word combinations emerge, emergence of narrative skills), since their age of acquisition is quite stable cross-linguistically for young children (Slobin, 1969). An adaptation of a screening tool principally targeting such milestones was judged to likely be adequate in the new language.

The Dutch parent questionnaire presented in Luinge et al. (2006) possessed all the required characteristics. Indeed, this parent questionnaire fulfilled Criteria 1 and 6, with items targeting vocabulary, syntax, narrative skills, and phonological development for both the expressive and receptive modalities. The items were selected under a "unitary dimension" view of language (Luinge et al., 2006, p. 924), according to which a child who has difficulty in one language domain is expected to have difficulty in other language domains, either concurrently or later. Following this theoretical viewpoint, different language domains were thus targeted in the parent questionnaire. It also fulfilled Criteria 2, 3, and 4: it is filled out by the parent, has a short administration and scoring time, and is aimed at children between 12 and 71 months. It originally contained 26 yes/no items, asking if the child says/comprehends/uses certain linguistics elements. Regarding Criterion 5, since only one questionnaire was found, any positive documentation of the tool's psychometric properties would be considered as adequate. Luinge et al. (2006) reported a cross-sectional study of 527 Dutch-speaking children between 12 and 72 months from four regions of the Netherlands, from a variety of city sizes with a roughly equal number of boys and girls per age group. The authors performed an item analysis to identify the most adequate items for each of the five age groups, leading to a final version with 14 items in total. A study with 98 participants that used the 14-item version of the questionnaire concluded that this parent questionnaire had a sensitivity of 94% and a specificity of 83% (Luinge, 2005), which are respectively considered as *good* and *fair* using the criteria proposed by Plante and Vance (1994). Although the validation of the questionnaire cannot be extended to any adapted version, it was hypothesized that the fact that the original version had documented validity increased the likelihood that an adapted version would also be valid. Finally, the questionnaire fulfilled Criterion 6, as the items were selected based on a literature review of language milestones of various English screening tools. Some items targeted skills that can be hypothesized to be relevant for any language, such as understanding two-word combinations and asking questions.

The original set of 26 items was kept for the adaptation for three reasons: (a) the differences between Dutch and French might influence which items are most adequate, (b) the difference between the two countries in terms of demographics and education might impact the value of the items, and (c) in the development of the original Dutch version, exclusion of items was performed before the questionnaire was tested on children with DLD. Thus, it is possible that some items from the original version would prove helpful to identify children with DLD or to characterize normal language development in French, even if they were not deemed necessary for characterizing normal language development in Dutch.

Adaptation procedures. The procedure to adapt the Dutch questionnaire to French involved four steps: (a) a translation and analysis of the original items to ensure their suitability in French; (b) a literature review to determine the necessity of adding items targeting language skills more specific to French; (c) whenever examples were provided in an item, a literature review was performed to select French examples representative of spontaneous utterances of francophone children; and (d) a review of the final items by native speakers of Québec French, not specialized in language development. The 29 items of the pilot version of the questionnaire are presented in Appendix A.

A direct translation of Dutch items into French was not always favoured because in some instances more casual vocabulary was considered preferable. For example, the direct translation of *speech* is *parole*, but this term is rarely used with this meaning by non-professionals. It was thus translated with *ce que dit votre enfant* [what your child says]. Once translated, each item was analyzed to make sure the target skill manifests in a similar way in both languages. In the Luinge et al. (2006) article, only the English version of the items is presented, hence the comparison will be made using the English. For example, the irregular plural marking, which is targeted in one item, implicates a similar modification of the noun in both languages. For example, *mouse* becomes *mice* in English, just as *cheval* [horse] becomes *chevaux* [horses] in French. In contrast, if the regular plural marking had been targeted, it would not have been judged equivalent. Indeed, English requires the application of a rule (i.e., adding an *-s* morpheme at the end of the word). However, in French, the regular noun plural is marked by a change in the determiner preceding the noun, the *-s* found in writing at the end of the noun is silent (e.g., *le chat* /ləʃa/ [the cat] vs. *les chats* /lɛʃa/ [cats]). Since all the items target language elements that manifest similarly in both languages, none were removed or modified based on this analysis.

The second step consisted of a literature review of French language development to determine if some important aspects of the French language, or some elements known to be particularly difficult for francophone children with DLD, should be targeted in new items. After a literature review of studies of French-speaking children with Specific Language Impairment (e.g., Elin Thordardottir & Namazi, 2007; Hamann et al., 2003; Jakubowicz, Nash, Rigaut, & Gérard, 1998) or *dysphasie* [dysphasia] (e.g., Roulet, 2007), three new items were created: gender agreement, the use of the object clitic, and subject omission.

The first of the additional items is related to the child's ability to make the correct gender agreement between a noun and its modifiers. In French, noun gender is marked on the accompanying determiner and, if present, adjective(s). The gender of the noun sometimes concurs with the referent's biological gender, as in *la fille* [$\text{the}_{\text{feminine}}$ $\text{girl}_{\text{feminine}}$], but most often the referent does not have a biological gender, as in *un crayon* [$\text{a}_{\text{masculine}}$ $\text{pencil}_{\text{masculine}}$]. Although gender agreement is an element acquired by children with typical development as early as 30–36 months for the *un/une* [a] contrast (Rondal, 2001), children with DLD between 6;11 and 11;3 have been reported to still make gender mistakes on the determiner or omit it (Roulet, 2007).

The second item targets the use of the object clitic, which is a pronoun placed between the subject and the verb used as a direct object complement. In a simpler syntactic structure, this complement would be placed directly after the verb, using a noun preceded by a determiner. For example, a typical sentence would be *Je veux la pomme* [I want the apple]. In a more complex syntactic structure where the direct object is pronominal, an object clitic is used and precedes the verb: *Je la veux* [I it want]. Evidence has been presented indicating that object clitics are particularly difficult for school-aged children with DLD in an elicitation task (Hamann et al., 2003; Jakubowicz et al., 1998). However, Elin Thordardottir and Namazi (2007) found in a study with children between 37 and 54 months that the object clitic did not seem to be an area of particular difficulty for children with DLD in their spontaneous language. Although these findings are somewhat conflicting, the inclusion of the object clitic was deemed to be warranted in a pilot version of the questionnaire.

The last item assesses the use of the subject, which is obligatory in most contexts in French. This item was selected because some authors have concluded that school-age Francophone children with language delays performed less well than age-matched peers in this respect in elicited production (Jakubowicz et al., 1998), although another study reported no difference between groups in spontaneous production (Hamann et al., 2003). Furthermore, Elin Thordardottir and Namazi (2007) found that children with DLD between 37 and 54 months omitted the first person singular pronoun *je* [I] more often than did age-matched peers. The use of sentences without subjects is related to an immature sentence construction, occurring when the child uses infinitive verbs without a subject such as *aller là* [go$_{\text{infinitive}}$ there] (Elin Thordardottir, 2005; Hamann et al., 2003). French-speaking children are reported to use syntactic subjects most of the time at 2 years of age (Parisse & Le Normand, 2001) and person marking of the verb has been shown to be productive at that age (Elin Thordardottir, 2005). It is thus expected that this item will be reported as acquired for children as young as 2 years of age.

The third step consisted of the selection of examples to help parents understand the items. A literature review was performed to select examples representative of typical utterances of young French-speaking children. Various sources were consulted, notably Bassano (2000), Parisse and Le Normand (2001), Hickman (1997), and Rondal (2001). The final step consisted of the revision of the 29 items by two adult native French-speakers without experience in linguistics or in speech-language therapy, to ensure that the items are easily understandable. Based on the comments, any required modifications (e.g., correction of typing mistakes, reformulation of some sentences, and addition of some examples) were performed.

Pilot testing of the parent questionnaire. A cross-sectional pilot study with a small number of typically developing monolingual participants from three age groups was conducted to determine (a) whether the pilot version is easily understood by parents (i.e., whether the wording of the items is adequate, whether the examples chosen are helpful to parents), (b) whether the items vary in difficulty, and (c) whether the questionnaire overall adequately captures different language skill levels in monolingual francophone children between 12 and 71 months.

Participants. The parents of 26 monolingual French-speakers participated in the study: nine children were between 12 and 23 months (1-year-old group), eight children were between 35 and 45 months (3-year-old group), and nine children were between 60 and 69 months (5-year-old group). All children had typical development (i.e., no diagnosis or parental concerns about the child's development or hearing). Maternal education level served as a measure of SES. Although this was not a goal in participant recruitment, all the participating parents were of relatively high SES: the mothers of 24 participants had attended university; the mothers of the remaining two children (one in the 1-year-old group, one in the 5-year-old group) had attended CEGEP. In the education system of the province of Québec, CEGEPs are postsecondary institutions providing a 2-year pre-university program, or a 3-year professional program; the first year of CEGEP is equivalent to Grade 12 in other Canadian provinces.

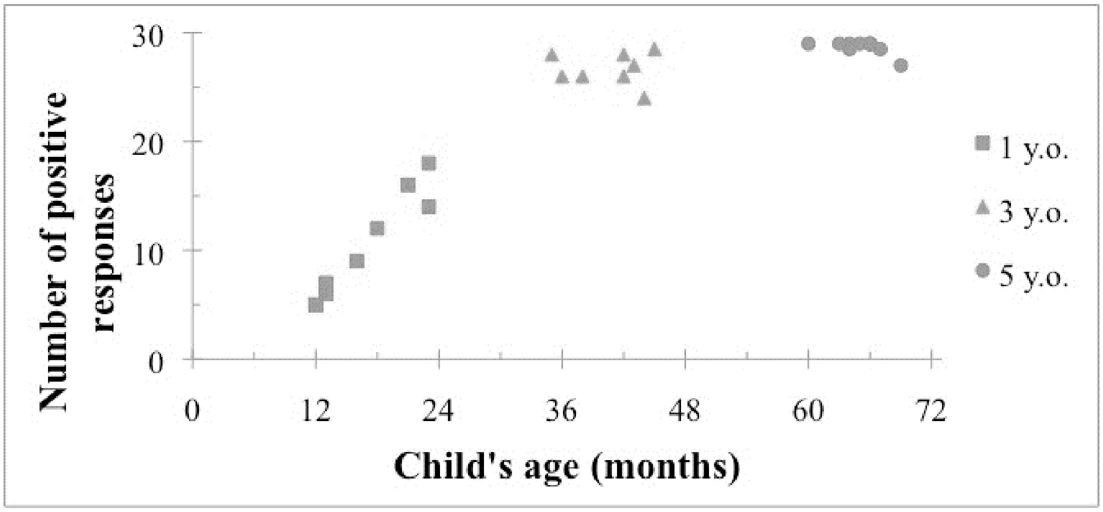
Procedures. The project was approved by the Institutional Review Board of McGill University (ethics approval #A05-E19-08B). The director of a Centre de la Petite Enfance sent a letter of invitation to participate in a study of French development to the parents of the children in the targeted age ranges; other participants were recruited by word of mouth. Parents who signed the consent form were asked to complete a background questionnaire about the child's development and other sociodemographic information, as well as the pilot version of the questionnaire with an added section for comments.

Scoring of the parent questionnaire was performed by assigning a score of 1 to items answered *yes* by the parent and a score of 0 to items answered *no* or without answer, leading to a maximum score of 29. In three cases across two items (namely Item 25 on the use of object clitic and Item 27 on the use of adult-like sentence complexity), parents provided written comments on the questionnaire's margin that made their answer both *yes* and *no*. For these cases, a score of 0.5 was credited.

Analyses and Results

Developmental sensitivity. The distribution of raw scores is shown in **Figure 1**. For the 1-year-old group, the scores increased systematically with age and no child reached the ceiling (i.e., none received a raw score of 28 or 29). Three of the eight children in the 3-year-old group were

Figure 1



The Raw Score of Each Child on the Pilot Version of the Parent Questionnaire as a Function of Age (max. score = 29).

at the ceiling. A ceiling effect was present for the 5-year-old group as most children (eight out of nine) reached the maximum score.

Item difficulty. In order to verify if the items varied in difficulty, the percentage of children per group who received a score of 1 was calculated for each item. The items were reordered based on a decreasing percentage of children receiving a score of 1 per age group (see **Table 1**). The easiest items ($n = 3$) were scored 1 for all children and the intermediate items ($n = 5$) were not scored as 1 for all children in the 3-year-old group, and were scored 0 for all children in the 1-year-old group. From these intermediate items, only one item was scored 0 by one participant in the 5-year-old group. There were no difficult items that would be scored 0 for most 3-year-old children and scored 1 by only a few of the 5-year-olds. The lack of sufficiently difficult items explains the presence of the ceiling effect observed for the 5-year-old group and that some 3-year-old children also reached the ceiling.

Parents' comments. Using the option provided at the end of the questionnaire, 17 parents indicated that it took less than 5 minutes to fill out the questionnaire, eight parents indicated that it took 5 to 10 minutes, and one parent took more than 15 minutes. Three parents indicated that the questionnaire was easy to fill out and not long to complete or both. Other comments were made regarding the formulation of some items; these are discussed in the next section.

Revision of the Questionnaire

Analyses of these results pinpointed several elements that could be improved in the pilot version of the questionnaire. Different types of changes were performed: The first required changes to the wording and ordering of the questions, the second consisted of the addition of items, and the last affected the answer choices provided to parents.

Wording and ordering of questions. A few parents requested a clarification of the word *généralement* [usually] used with some items; it was thus changed to *plus de 75% du temps* [more than 75% of the time]. Item 27, "*Est-ce que votre enfant parle comme un adulte, en ce qui a trait à la complexité des phrases?*" [Does your child talk like an adult, in terms of sentence complexity?], was annotated relatively frequently, with many parents making a comparison to peers rather than to adults. Therefore, the item was changed to a broad evaluation of the child's language skills compared to peers: "*Est-ce que vous considérez que votre enfant a un langage suffisamment développé, en comparaison aux autres enfants de son âge?*" [Do you consider that your child's language skills are sufficiently developed compared to other children of the same age?]. Regarding one item on intelligibility, a parent pointed out that she was almost the only one to understand her child's speech. Because it was judged pertinent to have parents report on their child's intelligibility to an unfamiliar person, an additional item targeting the need for a parent to act as an interpreter for his or her child was added.

Table 1

The Number of Children Receiving a Positive Response for Each Item on the Pilot Version of the Parent Questionnaire, With the Items Reordered by Difficulty

Item number	1-year-old (*n* = 9)	3-year-old (*n* = 8)	5-year-old (*n* = 9)	Total (*n* = 26)
1	**9**	**8**	**9**	26
3	**9**	**8**	**9**	26
9	**9**	**8**	**9**	26
2	8	**8**	**9**	25
4	8	**8**	**9**	25
11	8	**8**	**9**	25
7	7	**8**	**9**	24
6	5	**8**	**9**	22
5	5	**8**	**9**	22
22	4	**8**	**9**	21
8	4	**8**	**9**	21
26	3	**8**	**9**	20
14	2	**8**	**9**	19
10	2	**8**	**9**	19
15	2	**8**	**9**	19
16	2	**8**	**9**	19
19	1	**8**	8	17
13	1	**8**	**9**	18
17	1	**8**	**9**	18
28	1	**8**	**9**	18
18	0	**8**	**9**	17
12	0	**8**	**9**	17
24	0	**8**	**9**	17
29	0	**8**	**9**	17
20	0	6	**9**	15
25	0	6	*8.5*	14.5
27	0	*3.5*	*8.5*	12
21	0	3	**9**	12
23	0	3	8	11

Note. Bold numbers indicate the items with a positive score for all children in a group. Italics indicate the items with partial credit.

Finally, additional examples were provided where some annotations indicated that they would be helpful. The items were reordered to present the easiest items first (see **Table 1**).

Additional items. In order to address the ceiling effect, additional items targeting elements acquired between 3 and 5 years of age were selected and introduced. The first item targeted the contracted articles (e.g., *du* instead of *de le*), an element specific to French. Although no study investigating its use by children with DLD was found, the contracted article is reported to be acquired around the age of 4 years (Rondal, 2001); it is thus potentially useful to prevent some 3-year-old children from reaching the ceiling.

Eight additional items were added based on a review article by Sprenger-Charolles and Serniclaes (2003) on reading and writing acquisition in various languages, including French. These items targeted narration and metalinguistic skills. Narrative development starts around age 3–4 years and is considered relatively mature at around 7–9 years (for a review, see Kao, 2015; Veneziano, 2016). There is a growing literature documenting difficulties of children with DLD on this type of task (e.g., Boerma, Leseman, Timmermeister, Wijnen, & Blom, 2016; Elin Thordardottir et al., 2011; Rezzonico et al., 2015), although studies also indicate that narrative ability is not highly sensitive to the presence of language impairment (e.g., Elin Thordardottir et al., 2011). Metalinguistic skills, including phonological awareness, typically start to develop at 4 years of age (for French, see Lefebvre, Girard, Desrosiers, Trudeau, & Sutton, 2008).

Finally, pre-reading skills were also targeted based on the work of Justice, Bowles, and Skibbe (2006) on print knowledge of 3- to 5-year-old anglophone children. Although print knowledge has been shown not to be directly related to oral language skills (McGinty & Justice, 2009), many children with DLD are reported to have difficulties in this area (Boudreau & Hedberg, 1999; Gillam & Johnston, 1985). Since both English and French have an alphabetic writing system, it was considered likely that these findings could be generalized to French.

Answer choices. Some parents signaled some degree of uncertainty about their answers by writing additional information beside their responses or by expressing it orally to the first author. Thus, the *yes-no* answer choices were replaced by a Likert-like scale with four options that were nonetheless scored dichotomously. The answers *oui* [yes] and *il me semble* [I believe so] were both scored as 1 point; the answers *je ne crois pas* [I don't think so] and *non* [no] were both scored as 0 point. This change was meant to give parents a means to express some level of uncertainty in their answers.

These modifications led to the addition of 10 items, inserted in the questionnaire based on the reported age of acquisition, leading to a total of 39 items. These 39 items can be described as 1 general item asking if the parent thinks their child's language is sufficiently developed for his/her age, 8 items on expressive vocabulary, 2 items on receptive vocabulary, 4 items on expressive vocabulary/syntax, 3 items on receptive vocabulary/syntax, 4 items on expressive syntax, 4 items on narrative abilities, 2 items on language use/communication, 5 items on phonology/articulation, 3 items on meta-linguistic knowledge, and 3 items on pre-reading skills. This final version of the adaptation was named the *Milestones en français du Québec* (MilBec; Paul & Elin Thordardottir, 2010), with *Milestones* referring to the title of the article presenting the original Dutch questionnaire. The MilBec can be found in Appendix B.

Phase B: Preliminary Investigation of the MilBec's Psychometric Properties

The purpose of this second phase was to collect preliminary data, using a cross-sectional sample, about the developmental sensitivity of the MilBec for monolingual French-speaking children between 12 and 71 months and to assess its internal consistency. It was hypothesized that (a) there would be a linear relationship between MilBec score and age in months, when all participants were considered as a single group and (b) significant differences in mean scores would be found between successive age groups.

Method

Participants. The parents of 85 monolingual French-speakers (44 boys, 41 girls) between 12 and 71 months participated in the study, with 17 participants per 12-month age group. Based on the background questionnaire, children were included in the study if they were exposed to another language for no more than 5 hours per week, they had no previous diagnosis, and their parents had no concern about their development or hearing. Maternal education served as a measure of SES. One child born prematurely was excluded from the study because of the specific risks to language development associated with prematurity in the preschool years (van Noort-van der Spek, Franken, & Weisglas-Kuperus, 2012). Two children were excluded because of serious parental concerns regarding language development. All participants were living in the province of Québec, mainly in the Greater-Montréal area (n = 69). The data for 10 participants were extracted from an unpublished longitudinal study using the same version of the MilBec (Paul, 2016). For eight of these participants, the first data point was used; for the remaining two participants, the data point placing them in the 2-year-old group was used because this group had the lowest number of participants in the cross-sectional sample.

Procedures. The parents were invited to participate in a study on the validation of a parent questionnaire about language development via daycares, sports centres, school, and public billboards using e-mail, posters, or billboard postings. All parents signed the project's consent form, which was approved by the Ethics Board of the Centre de recherche interdisciplinaire en réadaptation du Montréal Métropolitain (ethics approval #CRIR-674-0112). After agreeing to participate, the parents filled out the MilBec and the background questionnaire using an on-line survey created with LimeSurvey (n = 39) or a paper-pencil version (n = 46), depending on their preference. The demographic questionnaire included questions about parental education level, the child's medical and developmental history, and language use at home. All statistical analyses were performed using the French version of IBM Statistic SPSS version 23.

Background variables. The background questionnaire of the cross-sectional study asked for maternal education in years. For the 10 participants from the unpublished longitudinal study, maternal education was available in terms of the highest level of education completed. For them, a high school diploma was considered as 11 years of education and university-level as 16 years. For each of the five groups, average age and maternal education are reported in **Table 2**. A one-way ANOVA showed that the five groups did not differ significantly on maternal education, $F(4, 79) = 0.30$, $p = .879$. A one-way ANOVA confirmed that the five groups differed significantly on age in months, $F(4, 80) = 447.22$, $p < .001$, with post hoc Tukey tests showing that each group differed significantly from the others (all $p < .001$).

Analyses and Results

Developmental sensitivity. The distribution of MilBec scores within each age group is shown using boxplots in **Figure 2**; the mean, standard deviation, and range of scores are presented in **Table 3**. The median and mean scores increase with age, with a greater group difference between the youngest groups than the oldest groups. The largest variability occurs at 2 years of age and the smallest at age 5. The mean and median are already high at age 3, with some children reaching the ceiling (i.e., a score of 38 or 39). For the 4-year-old group, an upper whisker of the boxplots is present and the standard deviation is similar to that of the younger groups, despite a high mean score and the fact that five children are at the ceiling. For the 5-year-old group, the lack of the upper part of the boxplot, the low standard deviation, the fact that 11 of the 18 participants are at the ceiling, and the lowest score in this group is 36, which is also rather close to the maximum score, indicate the presence of a ceiling effect.

An ANOVA was performed to test for an age effect. A significant Levene test ($p < .001$) indicated that the data violated the assumption of homogeneity of variance. Consequently, the Brown-Forsythe adjusted F test was used and showed a significant group difference, $F(4, 50.024) = 97.98$, $p < .001$. A post hoc Games-Howell test indicated that the 1-year-old group and the 2-year-old group were significantly different from all the other groups (all p values between <.001 and .008). The 3-year-old group was not significantly different from the 4-year-old group ($p = .771$), but was significantly different from the 5-year-old group ($p = .002$). The 4-year-old group was significantly different from the 5-year-old group ($p = .023$).

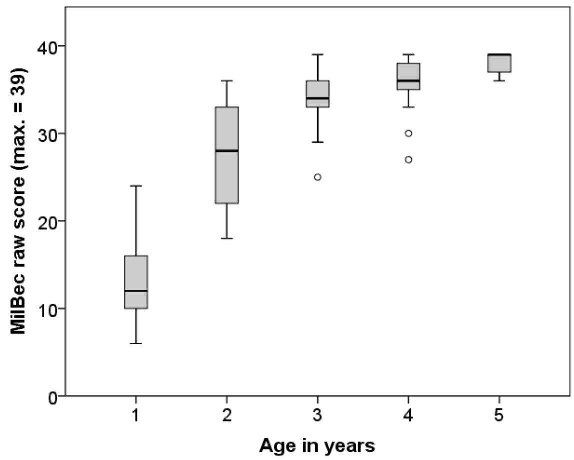
Visual inspection of the scatterplot showing the relation between age in months and MilBec scores indicated that the relationship was not linear (see **Figure 3**). Thus, a local regression (locally estimated scatterplot smoothing; LOESS) adjustment curve with the default Epanechnikov adjustment using 50% of the data points was performed on all the data since it can be used on empirical data to fit smooth curves without specifying an a priori relationship between the variables (Jacoby, 2000). Visual inspection of this LOESS curve indicated that the relationship between MilBec scores and age followed two distinct linear slopes—one for the younger children and one for the older children—with a relatively short period of transition around 40 months. The data were thus considered separately for the children between 12 and 39 months ($n = 42$; mean age = 26.5, $SD = 8.5$; mean MilBec

Table 2

Number of Participants, Age, and Maternal Education for Each Age Group

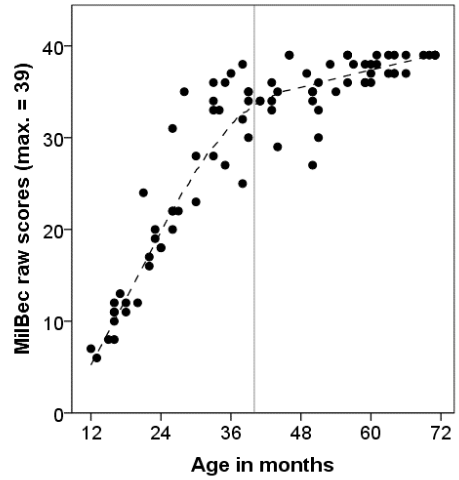
Group	*n*	Age (months) *M*	Age (months) *SD*	Age (months) Range	Maternal education (years) *M*	Maternal education (years) *SD*	Maternal education (years) Range
1-year-olds	17 (7 boys, 10 girls)	17.9	3.4	12–23	16.2	2.5	12–21
2-year-olds	17 (6 boys, 11 girls)	29.6	3.9	24–35	16.6	3.0	11–20
3-year-olds	17 (11 boys, 6 girls)	41.0	3.0	36–46	16.3	2.1	13–19
4-year-olds	17 (12 boys, 5 girls)	53.6	3.6	49–59	16.4	3.2	11–24
5-year-olds	17 (8 boys, 9 girls)	64.9	4.1	60–71	15.8	1.8	11–18

Figure 2



Boxplot of the *Milestones en français du Québec* (MilBec) Score per Age Group (max. score = 39). The median is shown as the line in the middle of the box, which itself indicates the range encompassing 50% of the scores; the whiskers show the range of the top and bottom 25% of the scores; the dots represent outliers.

Figure 3



Scatterplot of the *Milestones en français du Québec* (MilBec) Score as a Function of Age (max. score = 39). The dotted line indicates the LOESS adjustment curve. The vertical gray line at age 40 months indicates when the change in slope is judged to occur.

score = 22.6, *SD* = 10.0, range 6–38) and the children between 40 and 71 months (*n* = 43; mean age = 56.0, *SD* = 9.0; mean MilBec score = 36.4, *SD* = 2.9, range 27–39). Using Pearson correlations, the strength of the relationship between age and MilBec score was $r = .92$, $p < .001$, for the younger group and $r = .60$, $p < .001$, for the older group.

Internal consistency. A common way to measure the internal consistency of a scale is to use Cronbach's alpha (α), which reflects the average correlation between all the possible combinations of the two halves of the item list. A high internal consistency is considered evidence of construct validity. The internal consistency of the MilBec was calculated based on all 39 items, with a resulting α = .967; if any of these 39 items were to be removed, the new α varied between .968 and .965. The removal of any of the 39 items would thus not improve the internal consistency of the questionnaire, even if two items (namely Item 3 and 5) showed no variability (i.e., all participants received a score of 1). Because of the different relationship between age and MilBec score for younger and older children, Cronbach's alpha was also calculated for each subgroup, with a resulting α = .961 for children between 12 and 39 months and α = .763 for children between 40 and 71 months. The analysis also showed that for the older group, in addition to the two items previously mentioned, 13 items had no variability across children.

Discussion

The goal of this article was to present the steps that led to the creation of the MilBec, a new parent questionnaire that could eventually be used to identify children between 12 and 71 months at risk of having a developmental language disorder, as well as a preliminary investigation of its psychometric properties. The MilBec is an adaption of the Dutch parent questionnaire presented in Luinge et al. (2006) and includes 39 items targeting various language domains to mirror the heterogeneity of the manifestation of DLD (Bishop et al., 2017; Leonard, 2014). The items of the MilBec include an adaptation of the original items from Luinge et al. (2006), as well as additional items selected based on published research on the development of French by children with typical development and children with DLD.

The first phase of the article described the steps involved in the development and piloting of the questionnaire. The preliminary investigation of the psychometric properties of the MilBec reported in Phase 2 of the article focused mainly on the documentation of its developmental sensitivity and internal consistency for a group of monolingual French-speaking children between 12 and 71 months. The results on developmental sensitivity partly concurred with the hypotheses since scores increased with increasing age. However, the relationship

Table 3

Milestones en français du Québec (MilBec) Score for Each Age Group

Group	*n*	*M*	*SD*	Range
1-year-olds	17	12.8	4.9	6–24
2-year-olds	17	27.4	6.4	15–36
3-year-olds	17	34.1	3.6	25–39
4-year-olds	17	35.4	3.1	27–39
5-year-olds	17	38.1	1.1	36–39

was not linear across the entire age range, but instead indicated two subgroups with different slopes, with MilBec scores increasing linearly within each group. For children between 12 and 39 months there was a very strong correlation between age and MilBec score (r = .92) and for children between 40 and 71 months the correlation was strong (r = .60), despite a ceiling effect occurring for the 5-year-old group and the fact that some children in the 3- and 4-year-old groups also reached the maximum score. It is possible that the children's scores are generally high partly due to the high SES of their families since children from high SES tend to have higher language skills (Fernald, Marchman, & Weisleder, 2013; Hoff, 2006; Perkins, Finegood, & Swain, 2013). Another reason could be an insufficient number of sufficiently difficult items.

The second hypothesis, which stated that significant differences in mean score would be found between successive age groups, was also partially confirmed since the mean scores of each age group were significantly different from each other, with the exception of the 3- and 4-year-old groups which were not statistically different from each other. It is possible that the lack of a statistically significant difference between these groups is partially due to a lack of power, given the low sample size. It could also be related to the need for more advanced items. Whether this will render the MilBec inadequate as a language screening tool for children over the age of 40 months can only be established with the documentation of its diagnostic accuracy in a future study.

The internal consistency of the MilBec was found to be very high based on Cronbach's alpha (α = .967). Possible explanations for a very high Cronbach's alpha are a high number of items and the possibility that some of them are redundant and should be removed in a revised version of the questionnaire (Tavakol & Dennick, 2011). The ceiling effect observed for the 5-year-old group, and an already high performance of some children in the 3- and 4-year-old groups, may also contribute to the very high internal consistency. The analysis pointed to two items that might be considered for removal in a revised version of the MilBec, as all participants received a score of 1. However, this finding would have to be replicated in a larger group of children more representative of the population also assessing whether children with DLD may obtain a score of 0 on these items. While these considerations warrant further examination, the results indicate that the questionnaire has adequate internal consistency.

The results indicate that the MilBec is understood by parents and that it is sensitive to language development in French-speaking monolingual children. The change in the slope describing the linear relation between age and MilBec score around 40 months, as well as a much higher number of items without variability for children between 40 and 71 months and a ceiling effect for the 5-year-old group, suggest that scores should be interpreted differently for the two age groups. For children under 40 months, it might be adequate to transform the raw score into a z score, whereas for older children it might be most adequate to only consider whether the score is above or below the cutoff value. It is also possible that because of the ceiling effect observed in the 5-year-old group and the similarity in performance between the 3- and 4-year-old children, the MilBec may not be able to achieve adequate specificity for the older children (i.e., that even children with DLD would receive a high score). This could only be established in a future screening accuracy study comparing the performance of children with and without DLD.

Limitations

Among the limitations of this study is the small sample size. In the cross-sectional study, group sizes of 17 or more are adequate for this preliminary study, but a larger sample is clearly required for a better representation of the population and for increased statistical power. A second limitation of the sample is that the parents are all of relatively high SES. In a study with a small sample size, homogeneity in background variables is beneficial as it prevents the effect under test (here, the effect of age) to be overshadowed by other variables. At the same time, such homogeneity lessens the sample's representativeness of the more general population. Therefore, further larger scale study of the MilBec will need to include diverse SES levels.

Further, the possible effect of schooling on the children's success on some items should also be assessed for older children, particularly for the items that target metalinguistic skills and pre-reading knowledge, which are explicitly taught in kindergarten. Future analyses based on a larger sample size should also investigate the potential effect of gender on the children's performance. Indeed, early language development may be different between boys and girls, with a slight advantage for girls (Wallentin, 2009), particularly between 17 and 28 months, where the expressive skills of French-speaking girls were found to be slightly better on the MacArthur Communicative Development Inventories (Bouchard, Trudeau, Sutton, Boudreault, & Deneault, 2009).

Conclusion

The creation and validation process of a new assessment tool is long and requires the documentation of various elements, including the ease of use by the persons who will complete it, and documentation of its validity, reliability, and diagnostic accuracy. As more information on an assessment tool is available, decisions can be made about whether the tool is adequate for its purpose and whether it can be revised to improve its value as a clinical tool. The preliminary results reported here are promising. The MilBec is currently used in different research projects to further document its psychometric properties, notably with bilingual and monolingual children with and without DLD.

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Appendix A

Pilot Version of the Parent Questionnaire

		oui	non
1.	Est-ce que votre enfant produit une variété de sons qui ressemblent à des consonnes et des voyelles?	☐	☐
2.	Est-ce que votre enfant dit « maman » ou « papa »?	☐	☐
3.	Est-ce que votre enfant comprend la signification de « non »?	☐	☐
4.	Est-ce que votre enfant produit quelques mots (simplifiés ou non)? (p. ex. *ati* pour « partie »; *la* pour « lait »; *non*)	☐	☐
5.	Est-ce que votre enfant peut identifier une ou plusieurs parties du corps? (p. ex. en répondant à des questions du type « Où est ton nez? »)	☐	☐
6.	Est-ce que votre enfant dit environ 10 mots différents?	☐	☐
7.	Est-ce que votre enfant peut pointer certains objets que vous nommez?	☐	☐
8.	Est-ce que votre enfant peut combiner deux mots? (p. ex. *veux biberon; là bobo; dedans chien*)	☐	☐
9.	Est-ce que votre enfant comprend des demandes simples de deux mots? (p. ex. « viens manger »; « assis-toi »)	☐	☐
10.	Est-ce que votre enfant fait des suites de trois mots? (p. ex. *veut monter Grégoire; pas mettre ça; moi goûter fraises*)	☐	☐
11.	Est-ce que votre enfant comprend des phrases de trois-quatre mots? (p. ex. « touche pas à ça »; « sur la table »; « attends ton tour »)	☐	☐
12.	Est-ce que votre enfant fait des phrases complètes de trois ou quatre mots? (p. ex. *on dirait une fille; il criait tout le temps; raconte une histoire*)	☐	☐
13.	Est-ce que votre enfant nomme correctement certaines couleurs?	☐	☐
14.	Est-ce que votre enfant pose des questions?	☐	☐
15.	Est-ce que votre enfant utilise le bon ordre des mots dans ses phrases?	☐	☐
16.	Est-ce que votre enfant mentionne le sujet dans ses phrases, c'est-à-dire est-ce qu'il indique qui fait l'action? (p. ex. *Martin* dans « Martin va à la piscine »; *tu* dans « Tu viens ? »)	☐	☐
17.	Est-ce que votre enfant utilise des mots qui qualifient/décrivent d'autres mots? (p. ex. *grande* et *rouge* dans « grande maison rouge »)	☐	☐

Pilot Version of the Parent Questionnaire

	oui	non
18. Est-ce que votre enfant peut répéter une histoire en se basant sur des images?	☐	☐
19. Est-ce que votre enfant raconte spontanément des événements de sa journée? (p. ex. quelque chose qui est arrivé à la garderie)	☐	☐
20. Est-ce que votre enfant utilise le masculin et le féminin correctement la majorité du temps?	☐	☐
21. Est-ce que votre enfant utilise généralement le pluriel correctement? (p. ex. yeux/œil; chevaux/cheval)	☐	☐
22. Comprenez-vous environ la moitié (50%) de tout ce que votre enfant dit?	☐	☐
23. Est-ce que votre enfant utilise correctement les passés composés irréguliers? (p. ex. *couru; mis; pris*)	☐	☐
24. Est-ce que votre enfant fait de longues phrases avec plusieurs verbes? (p. ex. *Quand le soleil se couche, il fait noir; Maman dit tu dois venir*)	☐	☐
25. Est-ce que votre enfant remplace parfois le mot qui désigne un objet par un pronom? (p. ex. *la* dans « Je la veux », au lieu de dire « Je veux la pomme »)	☐	☐
26. Comprenez-vous environ les trois quarts (75%) de tout ce que votre enfant dit?	☐	☐
27. Est-ce que votre enfant parle comme un adulte, en ce qui a trait à la complexité des phrases?	☐	☐
28. Est-ce que votre enfant comprend des consignes à deux étapes ou plus? (p. ex. « Tu dois ranger tes jouets avant d'aller jouer dehors »)	☐	☐
29. Comprenez-vous la quasi-totalité (près de 100%) de tout ce que votre enfant dit?	☐	☐

Appendix B

« Milestones » en français du Québec (MilBec)

Pour dépister les difficultés langagières des enfants de 12 à 71 mois

Nom de l'enfant : ______________________ Genre : ☐ masculin ☐ féminin

Date de naissance (jj-mm-aaaa) : ______________ Âge (mois) : ______________

Complété le (jj-mm-aaaa) : ______________ Par : ☐ mère ☐ père ☐ autre ______________

Consignes : Indiquez « oui » si la réponse est vraie présentement ou l'était lorsque votre enfant était plus jeune.

Questionnaire – Merci de répondre à toutes les questions

Comme le même questionnaire est utilisé pour tous les enfants, il est normal que les enfants plus jeunes aient une majorité de réponses négatives.

	Question	oui	il me semble	je ne crois pas	non
1.	Est-ce que vous considérez que votre enfant a un langage suffisamment développé, en comparaison aux autres enfants de son âge?	☐	☐	☐	☐
2.	Est-ce que votre enfant produit, ou produisait quand il était petit, une variété de sons qui ressemblent à des consonnes et des voyelles?	☐	☐	☐	☐
3.	Est-ce que votre enfant comprend la signification de « non »?	☐	☐	☐	☐
4.	Est-ce que votre enfant comprend des consignes simples de deux mots? (p. ex. « viens manger »; « assis-toi »)	☐	☐	☐	☐
5.	Est-ce que votre enfant dit « maman » ou « papa »?	☐	☐	☐	☐
6.	Est-ce que votre enfant produit quelques mots (simplifiés ou non)? (p. ex. *ati* pour « partie »; *la* pour « lait »; *non*)	☐	☐	☐	☐
7.	Est-ce que votre enfant comprend des phrases de trois ou quatre mots? (p. ex. « touche pas à ça »; « sur la table »; « attends ton tour »)	☐	☐	☐	☐
8.	Est-ce que votre enfant vous montre du doigt les objets qui l'intéressent?	☐	☐	☐	☐
9.	Est-ce que votre enfant peut identifier une ou plusieurs parties du corps? (p. ex. répond à des questions du type « Où est ton nez? »)	☐	☐	☐	☐
10.	Est-ce que votre enfant dit environ 10 mots différents?	☐	☐	☐	☐
11.	Est-ce que votre enfant peut combiner deux mots? (p. ex. *veux biberon*; *là bobo*; *dedans chien*; *papa parti*)	☐	☐	☐	☐

Questionnaire – Merci de répondre à toutes les questions

Comme le même questionnaire est utilisé pour tous les enfants, il est normal que les enfants plus jeunes aient une majorité de réponses négatives.

		oui	il me semble	je ne crois pas	non
12.	Comprenez-vous environ la moitié (50%) de tout ce que votre enfant dit?	☐	☐	☐	☐
13.	Comprenez-vous environ les trois quarts (75%) de tout ce que votre enfant dit?	☐	☐	☐	☐
14.	Est-ce qu'il vous est inutile de « traduire » ce qu'a dit votre enfant pour qu'une personne non familière le comprenne, plus des trois quarts (75%) du temps?	☐	☐	☐	☐
15.	Est-ce que votre enfant fait des suites de trois mots? (p. ex. *veut monter Grégoire; pas mettre ça; moi goûter fraises*)	☐	☐	☐	☐
16.	Est-ce que votre enfant pose des questions (avec des phrases complètes ou non)? (p. ex. *Papa parti?; est où Maman?; pourquoi?*)	☐	☐	☐	☐
17.	Est-ce que votre enfant fait toujours ses phrases avec les mots dans le bon ordre?	☐	☐	☐	☐
18.	Est-ce que votre enfant mentionne le sujet dans ses phrases, c'est-à-dire est-ce qu'il indique qui fait l'action? (p. ex. *Martin* dans « Martin va à la piscine »; *tu* dans « Tu viens? »)	☐	☐	☐	☐
19.	Est-ce que votre enfant raconte spontanément des événements de sa journée? (p. ex. quelque chose qui est arrivé à la garderie)	☐	☐	☐	☐
20.	Est-ce que votre enfant nomme correctement certaines couleurs?	☐	☐	☐	☐
21.	Est-ce que votre enfant possède dans son vocabulaire trois mots ou plus qui qualifient ou décrivent d'autres mots? (p. ex. *grande* et *rouge* dans « grande maison rouge »; *très* dans « très vite »)	☐	☐	☐	☐
22.	Est-ce que votre enfant comprend des consignes à deux étapes ou plus? (p. ex. « Tu dois ranger tes jouets avant d'aller jouer dehors »)	☐	☐	☐	☐
23.	Est-ce que votre enfant fait des phrases complètes de trois ou quatre mots? (p. ex. *on dirait une fille; il criait tout le temps; raconte une histoire*)	☐	☐	☐	☐
24.	Est-ce que votre enfant peut répéter une histoire en se basant sur des images?	☐	☐	☐	☐
25.	Est-ce que votre enfant fait de longues phrases avec plusieurs verbes? (p. ex. *Quand le soleil se couche, il fait noir; Maman dit* : « *tu dois venir* »)	☐	☐	☐	☐
26.	Comprenez-vous la quasi-totalité (près de 100%) de ce que votre enfant dit?	☐	☐	☐	☐
27.	Est-ce que votre enfant utilise le masculin et le féminin correctement la majorité du temps? (p. ex. *la pomme; la gentille fille; un tapis; le beau chien*)	☐	☐	☐	☐

Questionnaire – Merci de répondre à toutes les questions

Comme le même questionnaire est utilisé pour tous les enfants, il est normal que les enfants plus jeunes aient une majorité de réponses négatives.

	oui	il me semble	je ne crois pas	non
28. Est-ce que votre enfant remplace parfois le mot qui désigne un objet par un pronom? (p. ex. *la* dans « Je la veux », au lieu de dire « Je veux la pomme »)	☐	☐	☐	☐
29. Est-ce que votre enfant utilise le pluriel correctement plus de 75% du temps? (p. ex. yeux/œil; corail/coraux)	☐	☐	☐	☐
30. Est-ce que votre enfant utilise correctement le passé composé des verbes irréguliers fréquemment utilisés? (p. ex. *couru; mis; pris*)	☐	☐	☐	☐
31. Est-ce que votre enfant utilise les articles contractés correctement plus de 75% du temps? (p. ex. *du* pour *de le; au* pour *à le*)	☐	☐	☐	☐
32. Est-ce que votre enfant commente parfois la similitude entre des mots liés par le sens? (p. ex. *la robe fleurie a des fleurs; la feuille est lignée parce qu'elle a des lignes*)	☐	☐	☐	☐
33. Est-ce que votre enfant est capable de trouver des mots qui riment? (p. ex. *moufette* va avec *toilette; chat* va avec *rat; tapis* va avec *souris*)	☐	☐	☐	☐
34. Est-ce que votre enfant est capable de trouver des mots commençant avec le même son? (p. ex. *part* va avec *petit; lapin* va avec *loupe; manteau* va avec *melon*)	☐	☐	☐	☐
35. Est-ce que votre enfant informe plus de 75% du temps du lieu et des personnes impliquées de manière suffisante, lorsqu'il raconte un événement de sa journée?	☐	☐	☐	☐
36. Est-ce que votre enfant indique clairement plus de 75% du temps dans quel ordre les événements se sont déroulés, lorsqu'il raconte une histoire?	☐	☐	☐	☐
37. Est-ce que votre enfant peut réciter l'alphabet sans erreur plus de 75% du temps?	☐	☐	☐	☐
38. Est-ce que votre enfant est capable de reconnaître plus de 3 mots écrits? (p. ex. son nom; papa; maman; marque de commerce)	☐	☐	☐	☐
39. Est-ce que votre enfant regarde un livre en le tenant à l'endroit, en commençant au début et en tournant les pages une à la fois, plus de 75% du temps?	☐	☐	☐	☐
Sous-totaux				

Nombre de « oui » et de « il me semble » ____________

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