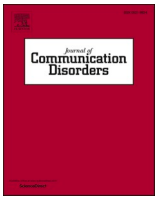
Journal of Communication Disorders 114 (2025) 106508

Contents lists available at ScienceDirect

Journal of Communication Disorders

journal homepage: www.elsevier.com/locate/jcomdis



Vocabulary and reading skills in adults with Prader-Willi syndrome

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ARTICLE INFO

Keywords:
Prader-Willi syndrome
Vocabulary
Reading
Lexical and phonological processes

ABSTRACT

Introduction: Prader-Willi syndrome (PWS) is a rare genetic condition associated with global intellectual impairment. While research has evidenced speech problems, little is known about reading, which is a critical language ability involved in communication. The aim of the present study was to investigate vocabulary and reading skills in adults with PWS.
Method: A total of 56 individuals (35 females, mean age = 33.64 years, range = 19–57 years) with PWS participated. Standardized paper and pencil tests were used to examine the level of vocabulary (LexTale-FR test) and reading performance (Alouette-R test). Two computerized tasks were also administered to assess the efficiency of lexical and phonological processes in reading (lexical and phonological decision tasks, taken from the ECCLA software). Performance was analyzed and compared with available norms on neurotypical adults and/or children.
Results: The results showed that adults with PWS had a low level of vocabulary (i.e., three to five standard deviations difference compared to neurotypical adults), poor reading skills (i.e., equivalent to the level of nine-year-olds), and less efficient lexical and non-lexical phonological processes.
Conclusion: The present data suggest a global impairment in vocabulary and reading skills in adults with PWS. These findings might help clinicians to better understand the language abilities of these patients.

1. Introduction

Prader-Willi syndrome (PWS) is one of the most common microdeletion syndromes (Cassidy, 1997). It is an imprinted condition caused by disruption of a several paternally inherited genes, particularly SNRPN, in the Prader-Willi critical region at chromosome location 15q11.2–15q13. It is associated with several behavioral and psychiatric characteristics that emerge with development (e.g.,

Author Note: This work was funded by a three-year PhD scholarship granted by the French Ministry of Higher Education and Research to Séverine Estival and a grant from the Prader-Willi Association France to Virginie Postal. It is part of a collaborative project between the Laboratory of Psychology of the University of Bordeaux (UR 4139) and the APHP (Assistance Publique - Hôpitaux de Paris) Hôpital Marin. We would like to thank all the participants of this study and the rehabilitation staff at the Hôpital Marin in Hendaye.

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https://doi.org/10.1016/j.jcomdis.2025.106508
Received 20 July 2024; Received in revised form 4 December 2024; Accepted 26 February 2025
Available online 27 February 2025

Hoybye, 2013). Extreme initial hypotonia and failure to thrive are followed by excessive appetite (hyperphagia) in early childhood and obsessive food-seeking in childhood, which can lead to morbid obesity in adulthood if not kept under control (Gunay-Aygun et al., 2001). Development is delayed with evidence of relative growth and sex hormone deficiency (short physical stature and impaired sexual development). Distinctive physical features are observed, such as small narrow hands, short broad feet, and specific facial features. A marked propensity to problematic behaviors is usually observed, such as temper outbursts, repetitive and ritualistic patterns and skin- picking (Whittington et al., 2004).

Although intellectual functioning varies across PWS individuals, global impairment in intellectual abilities is commonly reported (Copet et al., 2010; Curfs et al., 1991). Full-scale IQ score is usually low, ranging from 55 to 70 (Curfs et al., 1991; Dykens et al., 1992). While some studies reported no difference between verbal and performance IQ (e.g., Copet et al., 2010; Jauregi et al., 2007; Whittington et al., 2004), others reported that verbal IQ is lower than performance IQ (e.g., Cassidy et al., 1997; Dykens et al., 1992; Thompson et al., 1996). Delays in language development are usually observed and often exacerbated by difficulties in articulation, possibly linked to hypotonia (see Lewis, 2006). To date, research devoted to language abilities in adults with PWS is scarce and has mainly focused on the oral side of language performance, i.e., speech characteristics and narrative abilities. Overall, oral language abilities have been found to be significantly below chronological age, with speech production presenting more challenges than oral comprehension in individuals with PWS (Chen et al., 2010; Dimitropoulos et al., 2013, see Lewis et al., 2006 for a review). Little is known about the written language skills of adults with PWS, especially reading performance, which is a critical language skill involved in communication and the keystone supporting several cognitive abilities. By assessing the academic skills (i.e., arithmetic, reading/decoding, and reading/understanding) of 21 PWS individuals aged 13 to 46 with the K-ABC battery (Kaufman & Kaufman, 1983), Dykens et al. (1992) found a mean age-equivalent reading score of 8 years, 5 months in both decoding and understanding. These authors also showed that vocabulary, as assessed by the Peabody Picture Vocabulary Test-Revised (PPVT-R; Dunn & Dunn, 1981), was correlated with academic skills, with an age-equivalent score of 9 years, 2 months. Chronological age was not correlated with academic skills, suggesting that more learning opportunities with age did not lead to a higher reading performance. By examining cognitive (i.e., visual processing, academic achievement, and intelligence) and behavioral (i.e., maladaptive, adaptive, and obsessive-compulsive behaviors) differences between different subtypes of PWS, Butler et al. (2004) reported difficulties in letter-word identification and reading comprehension, as assessed by the Woodcock-Johnson Psycho-Educational Battery (Woodcock & Johnson, 1990). Although these studies conducted in English provided evidence of reading performance differences, the mechanisms leading to poor reading performance were not evaluated, as they were not designed to address this specific issue. Since word recognition and vocabulary are critical ingredients of proficient reading (e.g., see Murray, 2016), research on reading in individuals with PWS also needs to focus on vocabulary knowledge, as well as lexical and phonological skills.

Over the past ten years, the LexTALE test initially developed in English to assess language proficiency (Lemhöfer & Broersma, 2012) has been adapted in several languages such as French (i.e., LexTALE-FR, Brysbaert, 2013) and has become widely used to assess vocabulary skills in adults. In the LexTALE-FR test, the participants are asked to identify real words among a list of words and pseudowords (i.e., orthographically legal and pronounceable nonsense strings of letters) of various levels of difficulty. Some of the pseudowords are particularly difficult (e.g., pseudohomophones), so that the selection of words from pseudowords is subtil and taps into lexical quality (Brysbaert, 2013; Perfetti, 2007). The test consists of a nonspeeded lexical decision task that can be used either in a yes/no format, or in a format requesting only to choose the words known. This latter format, used in LexTALE-FR offers the advantage to be less demotivating for the participants that only know a few words (Brysbaert, 2013). Therefore, LexTALE-FR assesses the correct spelling of words, providing an index of the quality and retrieval of lexical representations stored in memory. Although initially proposed to assess language proficiency in bilinguals (Lemhöfer & Broersma, 2012), several authors suggested that LexTALE was also relevant to assess the size of the vocabulary in monolingual adults, whether in experimental or clinical settings (Brysbaert, 2013; Dujardin & Mathey, 2020).

The Alouette-R test (Lefavrais, 2005) is widely used to assess reading skills in French. In this test, participants are asked to read aloud a text with little meaning and several pitfalls, which facilitate the detection of reading difficulties. The drawings presented with the text can lead to mispronunciations if used by the reader. This strategy is often used by poor readers to facilitate understanding. Moreover, low-frequency words are inserted in the text where high-frequency ones could be expected, leading poor readers to make reading errors. While Alouette-R was initially used to detect dyslexia in children and young adults (e.g., Cavalli et al., 2017; Ecalle & Magnan, 2008), it has also been used to assess reading skills in adults (e.g., Dujardin & Mathey, 2020). Recent norms provided for the French language are available to classify individuals both for LexTALE-FR and Alouette-R scores (Dujardin et al., 2022). These norms have been developed from samples of native French speakers aged 18–26 years reporting no history of reading or oral language difficulty and having a mean education level of two years after the baccalaureate ($N = 410$ for the LexTALE-FR, and $N = 1231$ for the Alouette-R).

Since reading is a complex ability, it is important to examine the efficiency of cognitive processes underlying the processing of written words. Word recognition is an essential component of reading, which consists in matching printed information (i.e., written words) with the corresponding lexical representation stored in memory. In a dual-route approach to reading, two routes that provide direct and indirect pathways to the lexicon can be used to activate lexical units (see Coltheart et al., 2001). In the direct lexical route, direct matching is made between the letter units and the lexical unit. In the indirect route, letter units are translated into phonemic units through a conversion operation that implements rules of correspondence between graphemes and phonemes. Phonemic units are linked together to form a phonological representation of the printed word. This assembled phonological representation and the phonological representation already stored in memory are then matched. The indirect route is assumed to play a critical role in learning how to read, allowing beginners to read new words by matching new letter sequences with familiar phonological representations of words.

The efficiency of the two routes of reading can be assessed with speeded forced-choice tasks, such as lexical and phonological decision tasks from the ECCLA software for which normative data were developed from a sample of 160 adults with different levels of education (ranging from the end of primary school to the baccalaureate and over; Zagar et al., 1995). In the traditional yes/no lexical decision task, participants are typically asked to decide quickly whether a string of letters is a real word or not by pressing one of two identified buttons. This allows an examination of the speed and efficiency of accessing word representations stored in the lexicon, while untimed word selection such as that requested in the LextTALE provides single information on response accuracy with no time pressure. In the speeded lexical decision task, non-words (i.e., non-pronounceable letter strings) can also be used in addition to pseudowords (Zagar et al.,1995) which offers the advantage to make the yes/no task easier for the participants. Furthermore, the word characteristics are typically manipulated to examine effects designed to draw conclusions on cognitive processes of reading. In the ECCLA lexical decision task, the examination of how and to what extent the effects of word frequency and word regularity are differing from the norms enables conclusions to be drawn on the efficiency of the direct lexical route and on the degree of reliance on the indirect route as opposed to the direct one. The regularity effect is reputed to be particularly present with low-frequency words since high-frequency ones can be retrieved directly. In skilled readers, it has mainly been observed in reading aloud tasks owing to the use of the direct route (e.g., Waters et al., 1984). In young readers, in whom lexical units are still weak, less numerous and take longer to be activated, the regularity effect has also been noted with low-frequency words in the lexical decision task (Schmalz et al., 2013). This suggests that young readers rely heavily on the indirect route when performing a silent lexical decision task. A forced-choice phonological decision task can be used to assess further specific processes of the non-lexical indirect route (Zagar et al., 1995). In this task, the participants are requested to indicate quickly whether two visually-presented letter strings are pronounced in the same way (e.g., *cron* /kron/ and *kron* /kron/) or not (e.g., *clar* /klar/ and *clur* /klyr/). All letter strings are pseudowords, so reference to lexical units already stored in memory (i.e., real words) is not possible. This task provides therefore an index of the efficiency of grapheme-phoneme conversion and assembly operations.

The aim of this study was to characterize vocabulary and reading skills in a sample of French-speaking adults with PWS. To date, only a limited number of studies have explored reading performance in PWS. Although these studies have found reading difficulties associated with PWS, the underlying mechanisms behind these differences remain to be highlighted. Additionally, since the studies were conducted in English-speaking participants, reading performance in French-speaking adults with PWS remains unclear. The current study seeks to fill these gaps. First, we used standardized French tests, for which norms are available for neurotypical adults and/or children,[1] to examine breadth of vocabulary and general reading abilities. The aim was to quantify the difficulties we expected in both vocabulary and general reading scores in adults with PWS. Second, we administered computerized lexical and phonological tasks to investigate the efficiency of the direct route and indirect non-lexical route in reading. Norms defined from neurotypical adults were also used to quantify the difficulties observed in individuals with PWS. Based on previous studies suggesting reading differences in PWS (e.g., Butler et al., 2004; Dykens et al., 1992), we expected PWS adults to have difficulties in using the direct lexical route and to rely more heavily on the indirect route. Regarding the indirect route, we hypothesized that its use would be less efficient compared to neurotypical adults. However, based on the finding of an age-equivalent reading score of 8 years and 5 months in English patients (Dykens et al., 1992), the use of the indirect route was expected to be less affected than that of the direct route. In the lexical decision task, we predicted large effects of word frequency and regularity. Furthermore, in the phonological decision task, an effect of homography on homophone pseudowords was expected. Finally, we hypothesized a global difference in the various spheres of cognitive functioning in PWS adults by predicting significant relationships between the various measures of vocabulary and reading skills, as well as with indexes of intellectual functioning.

2. Method

2.1. Participants

A total of 56 individuals with PWS (35 females and 21 males) aged 19 to 57 years ($M = 33.64$, $SD = 7.93$) were recruited during a stay at the Hôpital Marin in Hendaye, a dedicated center for PWS patients in France. All participants and/or their caregivers gave their written informed consent after being told about the main aims of the study. All were native French speakers and reported having normal or corrected-to-normal vision. All had genetic confirmation of the PWS diagnosis, with 43 cases of deletion (76.79%), five cases of uniparental maternal disomy (8.93%), and eight other forms of genetic defect (imprinting center mutations or translocations, 14.28%). Intellectual abilities were evaluated in 45 patients with the third edition of the Wechsler Adult Intelligence Scale (WAIS-III, Wechsler, 1997). Overall cognitive efficiency was assessed with the MoCA (Montreal Cognitive Assessment, Nasreddine, 2005). Participants' characteristics are summarized in Table 1. The number of participants per test ranged from 45 to 56, depending on the test. This study was conducted in accordance with the ethical principles of the Declaration of Helsinki (2013). All ethical aspects of the recruitment process were governed by the French Health Code.

2.2. Materials and procedure

Two paper and pencil tests were used to assess vocabulary (LexTALE-FR; Brysbaert, 2013) and reading skills (Alouette-R; Lefavrais,

[1] For the different tests used in the present study, the comparison groups were neurotypical adults and/or children since norms on adults with intellectual disability are not available in French for these tests.

Table 1
Participants' Characteristics.

		Mean	SD
Age		33.64	7.93
% Females		62.50	–
% Right-handed		69.64	–
Years of education (n=39)		7.85	3.40
WAIS III (n=45)			
	Full-scale IQ	61.62	10.39
	Verbal IQ	63.04	11.16
	Performance IQ	63.40	11.55
MoCA (n=56)		20.50	4.20

Note. WAIS-III = Wechsler Adult Intelligence Scale-III; IQ = Intellectual Quotient, MoCA = Montreal Cognitive Assessment.

2005), and two computerized tasks were used to assess the efficiency of the lexical and phonological processes (ECCLA; Zagar et al., 1995).

2.2.2. *LexTALE-FR test*

In the LexTALE-FR test (Brysbaert, 2013), participants were asked to identify real words among 56 real French words and 28 pseudowords that resembled real words. The test was performed on a sheet of paper. There was no time limit. Two scores were computed to assess the participants' performance (see also Dujardin et al., 2022). First, the percentage of correct responses, which took into account both the correct selection of real words (W) and the correct deselection of pseudowords (PW), was computed using the following formula: (W + 2 x PW) /112. Second, a discrimination score (d'), which took into account both the correct responses on words (i.e., hit rates) and the incorrect responses on pseudowords (i.e., false alarms), was computed using the following formula: d' = Z (hit rates) - Z (false alarms).

2.2.3. *Alouette-R test*

In the Alouette-R test (Lefavrais, 2005), participants were instructed to read aloud a text of 265 words as quickly and accurately as possible. Maximum reading time was three minutes. Several measures were collected and/or computed: reading time (TL), number of words read (M), number of errors (E), number of words read correctly (i.e., C = M-E), index of precision (i.e., CM = (C/M) x 100), and index of efficiency (i.e., CTL = (C x 180) / TL). The latter index is widely used as it reports both the accuracy and speed of responses (e.g., Cavalli et al., 2018; Dujardin et al., 2022). Among these measures, we focused on the index of efficiency (CTL) and reading times (TL), since adult norms are available for these two measures (Dujardin et al., 2022).

2.2.4. *Lexical decision task*

The lexical decision task (ECCLA; Zagar et al., 1995) was programmed using the Superlab Pro software to assess lexical access processes. Each trial consisted of a sequence of two stimuli presented in isolation in the center of the screen of a personal computer. First, a fixation point appeared for 500 ms. Then, a string of lowercase letters appeared and remained on the screen until the participant responded or until 5000 ms had elapsed. Participants were instructed to decide as quickly and accurately as possible whether the target was a word or not by pressing one of two buttons on a keyboard. Visual feedback was provided when participants failed to respond or when 5000 ms had elapsed. In total, 96 stimuli were presented. Half were not real words, with 24 pseudowords (i.e., pronounceable strings of letters, e.g., *vacarin*) and 24 nonwords (i.e., unpronounceable strings of letters, e.g., *cfqhr*). The other half were real words differing by both word frequency and word regularity, so that four word conditions were distinguished: (1) 12 high-frequency regular words (e.g., *fromage [cheese]*), (2) 12 high-frequency irregular words (e.g., *monsieur [mister]*), (3) 12 low-frequency regular words (e.g., *haltère [dumbbell]*), and (4) 12 low-frequency irregular words (e.g., *quetsche [damson]*). Twenty-four practice trials were conducted before the experiment started. Accuracy and reaction times (measured from stimuli onset until the participant responded) were recorded. By comparing performance on low- and high-frequency words (i.e., word frequency effect), we obtained an index of the efficiency of the direct lexical route. By comparing performance on regular and irregular words (i.e., regularity effect), we obtained an estimate of the degree of reliance on the indirect route and on the direct route. Finally, the comparison between pseudoword and nonword performance was used to examine the reliance on grapheme-phoneme conversion operations.

2.2.5. *Forced-choice phonological decision task*

The computerized phonological decision task (ECCLA; Zagar et al., 1995) was programmed using the Superlab Pro software to assess the indirect non-lexical reading route. Each trial consisted of two sequences of stimuli. First, a fixation point appeared for 500 ms. Two pronounceable strings of letters (i.e., pseudowords) were then presented one below the other at the center of the screen of a personal computer. Participants were instructed to decide as quickly and accurately as possible whether the two pseudowords were pronounced similarly (i.e., homophones) or not by pressing one of two buttons on the keyboard. Visual feedback was provided when participants failed to respond or when 5000 ms had elapsed. In total, 64 trials (i.e., pairs of pseudowords) were presented so that 32 pairs could not be pronounced the same (i.e., non-homophones, e.g., *fagout-fugout*). The 32 remaining pairs could be pronounced the same way (i.e., homophones). Among them, 16 pairs had the same spelling (i.e., homograph homophones, e.g., *cefteu-cefteu*) and 16 had a different spelling (i.e., non-homograph homophones, e.g., *kron-cron*). Sixteen practice trials were conducted before the

experiment started. Accuracy and reaction times (measured from stimuli onset until the participant response) were recorded. Since pseudowords are not stored in the mental lexicon of participants, they necessarily require the use of the indirect non-lexical route to be read. The performance on homograph homophones was compared with that on non-homograph homophones to obtain an index of the efficiency of phonological processes.

3. Results

3.1. LexTALE-FR test

Mean scores and standard deviations computed on the 56 participants are presented in Table 2.

In comparison with the norms established among a French sample of young adults (Dujardin et al., 2022), the mean percentage of correct responses of individuals with PWS was very low (75.03 vs. 88.40). This performance was classified below the 5th centile (M = 80, range = 79–82). By computing z-scores, we estimated that the performance of PWS patients deviated significantly from the norms (M=-3.12, SD=2.32, p<.001). Similarly, mean d' was very low (-0.07), clearly below the mean score in the norms (2.65). This score was also classified below the 5th centile (M = 1.82, range = 1.71–1.92). Computation of z-scores also indicated that the vocabulary performance of PWS patients deviated significantly from the norms (M=-5.45, SD=1.91, p<.001).

3.2. Alouette test

Mean scores and SDs were computed on the 56 participants. Table 2 presents the comparison of the scores of PWS patients with norms on neurotypical adults (Dujardin et al., 2022), and Table 3 presents the comparison with norms established on children (Lefavrais, 2005).

In comparison with the norms from adults (see Table 2), the index of efficiency for PWS patients (i.e., CTL index) was very low (213.14 vs. 493.40), clearly below the 5th centile (M = 368, range = 355–378). By computing z-scores, we estimated that the performance of PWS patients deviated significantly from the norms (M=4.31, SD=1.06, p<.001). Similarly, reading times were very long (170.80 vs. 97.73), below the 5th centile (M = 126, range = 124–132). Computation of z-scores indicated that the performance of PWS patients deviated significantly from the norms (M=-3.39, SD=1.01, p<.001).

We also computed several indexes to compare scores with the norms established in a French sample of 398 children aged 6–16 (Lefavrais, 2005). Reading times, number of words read, number of errors, number of words read correctly, precision index, and speed index were reported. Table 3 presents the mean scores and SDs associated with the norms of Lefavrais (2005), indicating age-equivalent and class-equivalent scores. All indexes were coherent, pointing to an age-equivalent level of nine years old and a class-equivalent level of 3rd-to-4th grade.

3.3. Lexical decision task

Two out of the 56 participants declined to take part in the lexical decision task. Mean scores and SDs computed on the 54 remaining participants are presented in Table 4. Analyses were run using the JASP software (JASP Team, 2024).[2] Performance was then compared with norms on neurotypical adults (Zagar et al., 1995).

3.3.1. Analyses on word responses

Analyses of variance with word frequency and word regularity as within-subject factors were run on the mean reaction times (in ms) and the percentage of errors. No analyses were conducted on non-responses because of the very low rate (M = 1.31%).

The analysis of reaction times showed that the main effect of word frequency was significant, F (1, 52) = 81.75, p < .001, η^2 = 0.40. High-frequency words were recognized faster than low-frequency ones (1167 vs. 1527). The main effect of regularity was also significant, F (1, 52) = 27.02, p < .001, η^2 = 0.08. Regular words were recognized faster than irregular ones (1273 vs. 1421). Finally, no significant interaction was found between word frequency and word regularity, F < 1.

The analysis of the percentage of errors also showed a significant main effect of word frequency, F (1, 52) = 115.25, p < .001, η^2 = 0.44. High-frequency words caused fewer errors than low-frequency ones (3.89 vs. 28.37). The main effect of regularity was also significant, F (1, 52) = 54.09, p < .001, η^2 = 0.10. Regular words caused fewer errors than irregular ones (10.33 vs. 21.93). Finally, a significant interaction was found between word frequency and word regularity, F (1, 52) = 51.47, p < .001, η^2 = 0.08. Post-hoc comparisons with a Bonferroni correction indicated that the effect of regularity was significant with low-frequency words, t (52) = 10.27, p < .001, but not with high-frequency ones, t<1.

3.3.2. Analyses of pseudoword and non-word responses

Paired sample t-test analyses were run to compare non-word and pseudoword performance on mean reaction times (in ms) and percentage of errors. No analyses were conducted on non-responses because of the very low rate (M = 1.47%).

The analysis of reaction times showed that pseudowords were significantly longer to reject than non-words (1712 vs. 1024), t (53)

[2] Removing one outlier who differed by 3 SD from the mean of the group did not change the findings. We therefore decided to report the analyses conducted on the whole sample (N=54).

Table 2
Mean Scores (and SDs) on LexTALE-FR and Alouette-R Tests in PWS Patients and in Norms from Dujardin et al. (2022).

		Mean (and SD) in PWS patients	Mean (and SD) in Norms (Dujardin et al., 2022)
LexTALE-FR			
	% of correct responses	75.03 (9.95)	88.40 (4.29)
	d'	-0.07 (0.96)	2.65 (0.50)
Alouette-R			
	Index of efficiency	213.14 (83.15)	493.4 (82.62)
	Reading times	170.80 (17.99)	97.73 (16.95)

Table 3
Mean Scores (and SDs) for Each Index of the Alouette Test Computed for PWS Patients, and Age-equivalent and Class-equivalent Scores from Norms (Lefavrais, 2005).

	Mean (and SD) for PWS patients	Norms extracted from children (Lefavrais, 2005)	
		Age-equivalent score	Class-equivalent score
Index of efficiency	213.14 (83.15)	9 years	3rd to 4th Grade
Reading times	170.80 (17.99)	9 years	4th Grade
Number of words read	206.30 (55.78)	8–9 years	3rd to 4th Grade
Number of errors	10.71 (9.10)	10 years	5th Grade
Number of words read correctly	195.59 (58.31)	9 years	3rd to 4th Grade
Index of precision	93.85 (6.22)	9 years	3rd to 4th Grade

Table 4
Means (and SDs) of Reaction Times and Percentage of Errors in Lexical Decision Task.

		Reaction times (in ms)	Errors
Words			
	Regular high-frequency	1097 (438)	3.24 (5.47)
	Irregular high-frequency	1237 (411)	4.55 (7.15)
	Regular low-frequency	1449 (462)	17.43 (16.04)
	Irregular low-frequency	1605 (504)	39.32 (24.59)
Non-words		1024 (346)	1.93 (3.21)
Pseudowords		1712 (566)	22.40 (19.19)

$= 13.41$, $p < .001$. Effect size (as measured by Cohen's d) was d = 1.83, indicating a significant effect. The analysis of the percentage of errors showed that pseudowords caused more errors than non-words (22.40 vs. 1.93), $t\,(53) = 8.37$, $p < .001$. Effect size was d = 1.14, indicating a significant effect.

3.3.3. Comparison with norms

Norms on neurotypical adults (Zagar et al., 1995) provide several indexes corresponding to the effect of word regularity (i.e., regular vs. irregular words) as a function of word frequency (see Table 5). When considering the effect of regularity on high-frequency words, we found that the effect was clearly higher in PWS participants than in the norms, both in reaction times (M=140, SD=220 vs. M=34, SD=70 in the norms) and in terms of percentage of errors (M=4.55, SD=7.15 vs. M=2, SD=6 in the norms). This was also true for low-frequency words, for which the effect of regularity in PWS participants was higher than in the norms both in reaction times (M=173, SD=357 vs. M=98, SD=93 in the norms) and in terms of percentage of errors (M=39.32, SD=24.59 vs. M=34, SD=20 in the norms).

Table 5
Mean Scores (and SDs) on the Lexical Decision and Forced-Choice Phonological Decision Tasks in PWS Patients and in Norms from Zagar et al. (1995).

		Mean (and SD) in PWS patients	Mean (and SD) in Norms (Zagar et al., 1995)
Lexical Decision Task			
	Regularity effect on high-frequency words		
	RTs	140 (220)	34 (70)
	Errors	4.55 (7.15)	2 (6)
	Regularity effect on low-frequency words		
	RTs	173 (357)	98 (93)
	Errors	39.32 (24.59)	34 (20)
Forced-Choice Phonological Decision Task			
	%LHO	53.39 (30.01)	17 (16)

Note. %LHO = percentage of errors for non-homograph homophones

3.4. Forced-choice phonological decision task

Three out of the 56 participants declined to take part in the forced-choice phonological decision task. Mean scores of correct reaction times and percent errors for homophone pseudowords computed for 53 PWS participants are presented in Table 6. Non-homophone pseudowords used as distractors caused a mean of 25.20% of errors (SD = 21.42) and 3.36% of non-responses (SD = 5.74). Owing to the high error rate with non-homograph homophones (> 50%), reaction times of correct responses were not analyzed in this condition. Paired sample t-tests were run using the JASP software (JASP Team, 2024) on both the percentage of errors and the percentage of non-responses to examine the effect of homography (non-homographs vs. homographs) on homophone pseudowords. Performance was then compared with norms on neurotypical adults (Zagar et al., 1995).

3.4.1. Analyses of pseudoword responses

The analysis of the percentage of errors showed that the effect of homography of the homophones was significant, t (52) = 10.58, p < .001. Effect size (as measured by Cohen's d) was d = 1.45, indicating a significant effect. Non-homograph homophones yielded more errors than homograph homophones (53.39 vs. 8.73). The effect of homography was also significant in the analysis of the percentage of non-responses, t (52) = 3.54, p < .001. Effect size was medium (d = 0.49). Non-homograph homophones yielded more non-responses than homograph homophones (6.72 vs. 1.06).

3.4.2. Comparison with norms

We examined the %LHO index (ECCLA; Zagar et al., 1995), corresponding to the percentage of errors for non-homograph homophones (see Table 5). In comparison with the ECCLA norms, we found that %LHO was very high in PWS patients (M=53.39, SD=30.01 vs. M=17, SD=16). Computation of z-scores indicated that performance of PWS adults deviated significantly from the norms (M=2.27, SD=1.88, p<.01). No reaction time indexes on correct word responses were used, since they involved the non-homograph homophones condition in which the error rate was too high, as previously indicated.

3.5. Relationships between the different measures

To examine the relationships between the participants' vocabulary and reading skills and their intellectual characteristics (full-scale IQ, verbal IQ, performance IQ, MoCA scores), we conducted Pearson's bivariate correlational analyses. The results are presented in Table 7.

The various indexes drawn from the Alouette-R test correlated significantly with the indexes of intellectual functioning, suggesting that the lower the intellectual functioning index, the lower the reading performance. The index of efficiency of the LexTALE-FR test also correlated significantly with the various indexes of intellectual functioning and with the Alouette test scores. However, the d' index did not correlate significantly with most measures. The percentage of errors for non-homograph homophones in the forced-choice phonological decision task correlated significantly with the indexes of intellectual functioning on reading and vocabulary performance. In the lexical decision task, regularity effects on errors correlated significantly with most measures, particularly for low-frequency words. Overall, lexical and phonological indexes computed from reaction times did not correlate with each other, or with other measures of reading skills and intellectual functioning.

4. Discussion

Our sample of 56 French-speaking adults with PWS showed a poor vocabulary level and reading difficulties, as compared with norms in neurotypical adults. PWS adults also showed significant lexical and phonological effects, suggesting a global difficulty in both the direct lexical and indirect non-lexical routes of reading. Finally, vocabulary and reading measures were correlated with indexes of intellectual functioning. The results are discussed hereafter in relation with previous studies conducted on individuals with PWS.

First, PWS adults exhibited a low level of vocabulary in French. This finding is consistent with previous studies conducted in English using other vocabulary tests (e.g., Dykens et al., 1992; Lewis et al., 2002). The LexTALE-FR test (Brysbaert, 2013), which is a quick and easy-to-administer vocabulary test commonly used in neurotypical adults, thus appeared to be particularly suitable for examining vocabulary performance in individuals with globally reduced intellectual functioning. By using existing norms in neurotypical adults (Dujardin et al., 2022), we also observed that the vocabulary scores of PWS adults were three to five standard deviations below the means of neurotypical adults. This finding can be interpreted in terms of a smaller vocabulary size and a lower lexical quality (see Brysbaert, 2013; Perfetti, 2007) in our population. PWS adults also exhibited a low performance in both reading efficiency and reading times on the Alouette-R (Lefavrais, 2005). Compared with neurotypical adult norms (Dujardin et al., 2022), reading efficiency of PWS

Table 6
Means (and SDs) of Reaction Times, Percentage of Errors and Non-Responses for Pseudoword Homophones in Forced-Choice Phonological Decision Task.

Pseudoword conditions	Reaction times (in ms)	Errors	Non-responses
Homograph homophones	1937 (550.34)	8.73 (15.55)	1.06 (3.18)
Non-homograph homophones	- [1]	53.39 (30.01)	6.72 (11.75)

Note. [1] Owing to high rates of errors and non-responses, reaction times for correct responses were not presented and analyzed in this condition

Table 7
Pearson's Correlations Between Measures.

Variables	PIQ	VIQ	FSIQ	MoCA	Alouette-index of efficiency	Alouette-reading times	Lextale-% of correct responses	Lextale-d'	FCPDT-%LHO	LDT-RT-REGF	LDT-RT-REGR	LDT-ER-REGF
VIQ	.55***	–										
TIQ	.87***	.88***	–									
MoCA	.67***	.71***	.77***	–								
Alouette index of efficiency	.50***	.69***	.62***	.61***	–							
Alouette reading times	-0.47**	-0.58***	-0.53***	-0.51***	-0.83***	–						
Lextale % of correct responses	.37*	.67***	.58***	.67***	.72***	-0.52***	–					
Lextale d'	.32*	.21	.28	.32*	.15	-0.07	.05	–				
FCPDT-%LHO	-0.60***	-0.59***	-0.66***	-0.62***	-0.55***	.43**	-0.56***	-0.12	–			
LDT-RT-REGF	-0.003	-0.09	-0.06	.28*	-0.05	.03	-0.03	.27	-0.09	–		
LDT-RT-REGR	-0.16	-0.18	-0.18	-0.08	-0.13	.04	-0.04	-0.31*	.17	.13	–	
LDT-ER- REGF	-0.14	.003	-0.06	-0.13	-0.30*	.15	-0.30*	-0.04	.17	.008	.07	–
LDT-ER- REGR	-0.30*	-0.21	-0.26	-0.36**	-0.45***	.40**	-0.34*	-0.14	.45***	-0.08	.21	.19

Notes. PIQ = performance intellectual quotient, VIQ = verbal intellectual quotient, TIQ: full-scale intellectual quotient, MoCA= Montreal Cognitive Assessment score, FCPDT-%LHO = Forced-choice phonological decision task percentage of errors for non-homograph homophones, LDT-RT-REGF = lexical decision task effect of regularity for high-frequency words on reaction times, LDT-RT-REGR = lexical decision task effect of regularity for low-frequency words on reaction times, LDT-ER-REGF = lexical decision task effect of regularity for high-frequency words on errors, LDT- ER -REGR = lexical decision task effect of regularity for low-frequency words on errors.

* $p<.05$,
** $p<.01$,
*** $p<.001$

adults were three standard deviations below the norms and reading times were four standard deviations above the norms, indicating significant differences in these abilities. This is in line with previous data obtained with a battery of tests in English (Butler et al., 2004), so the Alouette-R is useful for assessing reading skills in adults with low intellectual functioning. Interestingly, as norms have also been established for French children (Lefavrais, 2005), we were able to estimate an age-equivalent level reached by the patients for several reading indexes. All the indexes pointed towards an age-equivalent level of 9 years old and a class-equivalent level of 3rd-to-4th grade in French education. Importantly, this age-equivalent reading level is quite similar to what was found by Dykens et al. (1992), even though the level we obtained was a little higher (i.e., 9 years old vs. 8 years, 5 months old) which may be due to the different assessment tools used and the age range of the participants. This small difference might also be due to language differences, since English orthography can be considered deeper than French orthography (e.g., Seymour et al., 2003). Further studies should be conducted to explore potential language differences. Overall, adults with PWS exhibit poor vocabulary and reading performance, which extends the previous data showing language difficulties in oral production to written language processing.

In addition to the low performance observed in vocabulary and reading tests, we also investigated the use of direct lexical and indirect non-lexical routes of reading (Coltheart et al., 2001). In the lexical decision task, we observed a large effect of word frequency, suggesting difficulties in lexical access and the less efficient use of the lexical route in individuals with PWS. Word frequency is considered as an index of greater efficiency of lexical access, more salient representations of words in memory, and greater availability of representations within individuals' vocabulary (Monsell, 1991). Significant regularity effects and difficulties in rejecting pseudowords rather than nonwords were also observed in PWS adults, providing evidence that the latter tend to rely heavily on the non-lexical indirect route and to use grapheme-phoneme conversion operations to process strings of letters. Finally, the forced-choice phonological decision task was useful to assess separately the efficiency of the processes underlying the use of the non-lexical indirect route, as only pseudowords were used as material. We observed that PWS individuals exhibited a large effect of homography on homophone pseudowords, with more than two standard deviations from the norms when processing non-homograph homophones. These findings suggest significant difficulties in performing phoneme-to-grapheme conversions. In a dual-route theoretical framework of reading (Coltheart et al., 2001), these findings and those of the lexical decision task suggest that the PWS individuals in this study preferentially used the indirect route of reading, even though conversion operations were not fully efficient. We also found relationships between the various measures of vocabulary and reading skills, as well as with indexes of intellectual functioning, supporting the hypothesis of a global weakness in the various spheres of cognitive functioning in these individuals. This provides further evidence of the global impairment of intellectual abilities commonly reported in PWS (e.g., Copet et al., 2010; Curfs et al., 1991). However, since the normative groups used for the various tests here consisted of neurotypical adults and/or children, rather than samples of adults with intellectual disabilities, further studies are needed to clarify the specific contribution of PWS in reading abilities independently of intellectual functioning. Interestingly, in the computerized tasks, errors correlated with the other language measures whereas reaction times did not. As the latter are particularly high in such tasks performed by PWS individuals, they do not seem to be a relevant indicator. In contrast, errors appeared to be a good indicator for investigating the efficiency of the direct lexical and indirect non-lexical routes. Finally, it is worth noting that left-handed individuals are over-represented in the present sample (about 30%) as compared to the general population (about 10%, see Papadatou-Pastou et al., 2020). A similar proportion has already been reported in a study on the same source population (Jauregi et al., 2007), which raises questions concerning the possible over-representation of left-handed in PWS (as already reported for several neuropsychiatric and neurodevelopmental conditions, such as schizophrenia, autism, and intellectual disability, e.g., see Papadatou-Pastou & Tomprou, 2015). Because of the known role of hemispheric lateralization underpinning both handedness and language abilities, further studies should be designed to address this issue in PWS.

In conclusion, this study contributes to a better understanding of vocabulary and reading performance in French-speaking PWS adults by providing further evidence of differences in vocabulary and reading skills. Additionally, the present findings demonstrate the effectiveness of the LexTALE-FR (Brysbaert, 2013) and Alouette (Lefavrais, 2005) tests for assessing vocabulary and reading performance in French-speaking PWS adults. These tests, commonly used in French-speaking neurotypical populations, therefore appear to be particularly well-suited for practitioners examining patients with low intellectual functioning. Investigation of the functioning of the direct and indirect routes of reading with speeded lexical and phonological decision tasks (Zagar et al., 1995) indicates a general difficulty such that not only the lexical mechanisms are not automatized but also the non-lexical phonological operations are not fully efficient. Given the difficulties in grapheme-to-phoneme conversion in adults with PWS, future research should also assess other abilities deemed necessary to achieve automatic word recognition, such as phonological awareness and decoding (e.g., Murray, 2016). Studies focusing on learning to read in children with PWS should also provide additional information on the reading challenges experienced by adults with PWS. Although further research is necessary to characterize these difficulties in adults, the present findings should encourage researchers and clinicians to pay particular attention to activities that call upon reading abilities and to develop appropriate interventions for individuals with PWS. In particular, the finding that vocabulary levels are low and that difficult words (e.g., low-frequency, irregular words) are hard to identify for adults with PWS suggests that researchers and clinicians should carefully consider the selection of words used in tasks assessing cognitive functioning and in interventions specifically designed for this population. Adapting reading materials based on the age-equivalent reading level identified here (i.e., 9 years old) could help facilitate the implementation of tasks and activities that require reading for adults with PWS.

CRediT authorship contribution statement

Christelle Robert: Conceptualization, Methodology, Software, Formal analysis, Visualization, Writing – original draft, Writing – review & editing. **Séverine Estival:** Investigation, Data curation. **Virginie Postal:** Project administration, Funding acquisition, Supervision, Writing – original draft. **Virginie Laurier:** Resources. **Fabien Mourre:** Resources. **Julie Tricot:** Resources, Investigation.

Stéphanie Mathey: Conceptualization, Methodology, Supervision, Visualization, Writing – original draft, Writing – review & editing.

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