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Factors explaining deficits in reading acquisition:

The case of Williams syndrome

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21 **ABSTRACT**

22

23 **Purpose:** Multiple factors impact reading acquisition in individuals with reading disability,
24 including genetic disorders such as Williams syndrome (WS). Despite a relative strength in
25 oral language, individuals with WS usually have an intellectual disability and tend to display
26 deficits in areas associated with reading. There is substantial variability in their reading skills.
27 While some authors have postulated that phonological deficits are at the source of their
28 reading deficits, others have suggested that they can be attributed to visuospatial deficits. The
29 present study was the first to undertake an in-depth exploration of reading skills among
30 French-speaking children and adults with WS. We tested the assumption that some factors
31 influence performance on single-word identification among individuals with WS, with a focus
32 on the roles of phonological awareness and visuospatial skills. **Method:** Participants were 29
33 French-speaking adults with WS and 192 controls matched for nonverbal mental age and
34 reading level. We administered tests assessing reading (decoding and word recognition),
35 vocabulary (expressive and receptive), phonological and visuospatial skills. We also
36 controlled for chronological age and nonverbal reasoning. **Results:** Phonemic awareness was
37 the most predictive factor of single-word identification in the WS group. Visuospatial skills
38 also contributed, but not more nor beyond other factors. More broadly, reasoning skills may
39 also have accounted for the variability in single-word identification in WS, but this was not
40 the case for either chronological age or vocabulary. **Conclusions:** There is considerable
41 heterogeneity among adults with WS, who may be either readers or prereaders. Similar
42 profiles identified among individuals with other specific learning disabilities suggest that high
43 reading variability is not specific to the neuropsychological profile of WS. We discuss a
44 multidimensional approach to the factors involved in reading deficits in WS.

45

46 **Keywords:** reading, multidimensional approach, phonological skills, visuospatial skills,
47 Williams syndrome

48

49 **1. Introduction**

50 Reading is a daily necessity for individuals who want to remain informed or entertained,
51 or who want to use modern technologies for social and communication purposes (*functional*
52 *literacy*; see World Literacy Foundation, 2015). Learning to read can be difficult for
53 individuals with intellectual disability, as is the case in genetic disorders such as Williams
54 syndrome (WS). However, studies on French-speaking individuals with intellectual disability
55 are still rare, even though learning to read is more complex for deep orthographies such as
56 French. *Deep orthographies* (e.g., French and English) are characterized by substantial
57 inconsistencies in the relationships between graphemes and phonemes, whereas shallow
58 orthographies (e.g., Spanish, Italian, and Hungarian) are characterized by more consistent
59 relationships (for a review, see Castles et al., 2018). French contains orthographic
60 inconsistencies and complexities, including multiletter graphemes (around 130 graphemes for
61 37 phonemes), context-dependent rules, irregularities, and morphological effects, but a simple
62 syllabic structure (Sprenger-Charolles & Ziegler, 2019). Orthographic depth affects the time it
63 takes to learn spelling-to-sound mapping (for dyslexia, see Paulesu et al., 2001), but not the
64 factors underlying reading skills (similar across European languages) (Caravolas et al., 2012).

65 Studying reading skills and their acquisition in French-speaking individuals with WS
66 could help them to gain equal educational and learning opportunities in our society. Although
67 the purpose of reading is ultimately to understand what is being read, the present study
68 focused on single-word reading, which can be regarded as a foundation skill for other
69 abilities, including reading comprehension (Sprenger-Charolles & Ziegler, 2019). As word
70 identification is a crucial step in reading acquisition, it is important to verify that it is fully
71 mastered before examining the higher-level processing required for comprehension.

72

73 *1.1. Cognitive and language profile in Williams syndrome*

74 WS is a rare neurodevelopmental disorder with an estimated prevalence of 1 in
75 7500-20 000, caused by the deletion of 16-28 genes on chromosome 7 (7q11.23) (Scherer &
76 Osborne, 2007; Strømme et al., 2002). Researchers have reported mild-to-moderate
77 intellectual disability in most individuals with WS, with an average intellectual quotient (IQ)
78 between 50 and 60 (for a review, see Martens et al., 2008). The cognitive profile of a typical
79 individual with WS is very heterogeneous (for a review, see Pezzino et al., 2017): language
80 production, receptive vocabulary and face recognition skills are considered to be relatively
81 preserved, whereas phonological processing (including phonological awareness¹ and
82 auditory-verbal memory) and visuospatial processes (including perceptual abilities,
83 visuoconstructive abilities, visuospatial memory) are severely affected (Dessaiegn et al.,
84 2013; Menghini et al., 2010; Pezzino et al., 2017; for neuroimaging studies, see Landau et al.,
85 2006). In this section, we focus on language skills and visuospatial skills, which are of
86 particular importance for the acquisition of reading.

87 *1.1.1. Language skills*

88 Vocabulary is not totally preserved in WS: performance is higher for receptive
89 vocabulary than for expressive vocabulary, with 86% of individuals with WS below the 10th
90 percentile in expressive vocabulary (Karmiloff-Smith et al., 1997; Laing et al., 2001; Martens
91 et al., 2008; Mervis, 2009; Monnery et al., 2002; Ypsilanti et al., 2005). More broadly, some
92 language skills in individuals with WS have been observed to follow the same pattern found
93 in typical development, but with a delayed developmental trajectory². These include
94 lexicosemantic skills, word fluency, syntax, morphosyntax, and semantic skills (Karmiloff-

¹ Phonological awareness (or metaphonological) is a meta-cognitive skill allows one to attend to, discriminate, remember, and manipulate sounds at the sentence, word and oral units (syllable and phonemes) level.

² This notion is developed in the neuroconstructivism approach of Karmiloff-Smith (1998), whereby some skills may be relatively preserved despite atypical development. When performance of individuals with WS is similar to or better than that of individuals matched on nonverbal mental age, it can be concluded that the development of the relevant skills is delayed but not atypical. In other words, development can show a typical pattern, but with a level equivalent to that of a child with a lower chronological age but the same mental age.

95 Smith et al., 1997; Martens et al., 2008). Other skills demonstrate an atypical pattern of
96 development, including phonological processing, verbal fluency, morphology, semantic
97 integration, and pragmatic skills (for French speakers, see Monnery et al., 2002; for Italian
98 speakers, see Bello et al., 2004; Burani et al., 2006; Vicari et al., 2004; for English speakers,
99 see Mervis, 2009; Perovic & Wexler, 2007; Ring & Clahsen, 2005; Stojanovik, 2006). These
100 early linguistic specificities contribute to a reduced lexical repertoire (Martens et al., 2008;
101 Perovic & Wexler, 2007; for the dysphasia profile, see Lahey & Edwards 1999).

102 Even within impaired language abilities, individuals with WS can display major
103 dissociations. For example, the literature on WS systematically highlights weaknesses in
104 phonological processing. However, in the case of phonological awareness, syllable awareness
105 is intact in individuals with WS (except for syllable suppression), and rime performance is
106 comparable to that of chronological age-matched peers, whereas phonemic awareness is
107 poorer (Garayzábal & Cuetos, 2008; Menghini et al., 2004). The source of these deficits is
108 unclear. Phonological deficits have been attributed to auditory-verbal memory deficits,
109 impeding phonological unit manipulation and phonological coding (using the phonological
110 loop) (Garayzabal & Cuetos Vega 2008; Karmiloff-Smith et al., 1997; Levy et al., 2003;
111 Majerus et al., 2003; Menghini et al., 2010; Rhodes et al., 2011; Temple, 2003; for a review,
112 see Majerus, 2019). Some studies point to an atypical development of the language system in
113 WS, with a detrimental effect of phonological variables on lexicosemantic variables,
114 potentially related to abnormal auditory-perceptual processing (Majerus et al., 2011). Other
115 studies also suggest that the phonological deficits could be explained by atypical development
116 of prosody (all oral features of verbal expression; see Catterall et al., 2006) or an excessive
117 cognitive load in phonological tasks (Cupples & Iacono, 2002; Levy et al., 2003).

118 *1.1.2. Visuospatial skills*

119 Regarding visuospatial skills, studies report the preservation of visuoperceptual skills
120 requiring intermediate or late perceptual processing (shape, object and face recognition; for
121 reviews, see Heiz, 2019). Deficits in early perceptual processing (line orientation judgement)
122 and visuoconstructive skills (copying, drawing and constructing cube patterns; Heiz, 2019;
123 Martens et al., 2008) seem to stem from difficulty with simultaneous local and global
124 processing (i.e. processing of details versus the broader pattern; Farran et al., 2003; Heiz &
125 Barisnikov, 2016), as well as from difficulty with mental manipulation of visuospatial
126 representations (as involved for example in the block design task; see Farran et al., 2003; for
127 neuroimaging studies, see Landau et al., 2006; Rondan et al., 2008). Visuomotor integration
128 skills demonstrates a typical pattern of development, but with a delayed trajectory, marked by
129 deficits in visuomotor perceptual integration and motor coordination in graphic production
130 tasks (Heiz, 2019; Heiz & Barisnikov, 2016; Wuang & Tsai, 2017). Individuals with WS also
131 have visuospatial working memory deficits (difficulty with maintaining and recalling serial
132 information; for a review, see Majerus, 2018, 2019). However, studies suggest more
133 difficulties with spatial structuring and visual exploration than with short-term storage
134 (Rhodes et al., 2011). Hoffman et al. (2003) described it as a *cascade effect* combining
135 executive deficits. More broadly, the nature of the task may affect the choice of local/global
136 processing and help to explain the visuospatial performance of individuals with WS (D'Souza
137 et al., 2016).

138

139 ***1.2. Reading ability in Williams syndrome***

140 Reading comprehension, the overarching goal of all reading activities, is the
141 multiplicative product of written word recognition (which requires explicit teaching before it
142 is automatized) and verbal comprehension (a natural process requiring no explicit teaching;
143 for a review, see Sprenger-Charolles & Ziegler, 2019). By the time they start learning to read,

144 learners already have relatively sophisticated spoken-language skills, including knowledge of
145 the meaning of many spoken words (Casalis et al., 2015). Written word recognition, however,
146 is another matter.

147 *Lexical quality* (the quality of stored mental representations specifying the form and
148 meaning of each word) is determined mainly by exposure to printed words (Perfetti, 2007). It
149 affects the development of orthographic representations and, consequently, single-word and
150 sentence reading performance (Casalis et al., 2015). If lexical quality is low, some of the
151 reader's limited cognitive resources have to be directed to word recognition (a low-level
152 process), meaning that comprehension (a high-level process) is compromised (for a review,
153 see Castles et al., 2018). The act of decoding words provides an opportunity to acquire
154 orthographic knowledge, which remains available for future encounters with the word (see
155 *self-teaching hypothesis*; Share, 2004). Once the alphabetic code (phoneme-to-grapheme
156 transcription) has been learned and understood, prereaders (i.e., individuals who are beginning
157 to learn how to read, and exhibit some basic decoding skills) can decode most printed words,
158 enabling them to access their pronunciation and vocabulary, and hence their meaning, if the
159 words are familiar in their oral form (Ehri, 2017; for a review, see Castles et al., 2018). As a
160 result of the self-teaching process during independent reading, less attention has to be paid to
161 the words' orthographic and phonological characteristics, enabling individuals to focus on
162 their meaning (Sprenger-Charolles & Ziegler, 2019).

163 In the case of WS, studies have shown delayed reading development from the early
164 stages onwards, compared with typically developing children matched on reading age (Steele
165 et al., 2013). The mean reading age of adult individuals with WS ranges from 6 to 8 years
166 (Laing et al., 2001; Levy et al., 2003; Udwin et al., 1996). The results of most studies of
167 reading skills in WS are consistent with the dual-route theory, which distinguishes between
168 two cognitive processes: visual word recognition and reading aloud (for computational

169 models, see Coltheart et al., 2001; Perry et al., 2010). The first route involves decoding words
170 by translating a word's spelling into its sound, thereby accessing its meaning. This makes it
171 possible for new and unfamiliar words to be read, based on alphabetic decoding. The second
172 route involves word recognition by gaining access to meaning directly from the spelling,
173 without having to decode its phonology. This allows for the reading of familiar words, where
174 there is direct access to meaning.

175 Research findings on the abilities involved in reading skills in WS seem to depend on
176 which language system is being studied. In the case of shallow orthographies, studies have
177 highlighted decoding deficits in Italian speakers, compared with mental and lexical age-
178 matched controls (Barca et al., 2010; Menghini et al., 2004), and word recognition deficits in
179 Spanish speakers, compared with mental age-matched controls (Garayzábal & Cuetos, 2008).
180 By contrast, in deep orthographies such as English, research has highlighted deficits in both
181 routes (Conners, 2003; Laing et al., 2001; Levy et al., 2003; Temple, 2003). Given this
182 variability in findings for WS, single-case studies and case series form an important aspect of
183 the evidence base in reading. For instance, a single-case study (Temple, 2003) of a 13-year-
184 old English-speaking girl with WS indicated both an inability to read pseudowords (PW) and
185 nonwords, highlighting a difficulty in word decoding, and semantic errors when reading
186 frequent words, highlighting a difficulty in word recognition. It has been suggested that the
187 reading profile in this deep orthographic system is similar to that observed in profound
188 dyslexia. Barca et al. (2010) reported the case of an Italian-speaking girl with WS aged 13
189 years and 8 months. In this shallow orthographic system, the teenager suffered a relative
190 weakness in decoding words, compared with recognizing words. In contrast to these cases
191 with decoding difficulties, Garayzábal and Cuetos (2008) reported that 12 Spanish-speaking
192 children with WS (mean age: 12 years and 5 months) performed similarly to mental age-
193 matched controls on nonword reading (word decoding), whereas their word reading (word

194 recognition) was impaired. A study of two English-speaking teenagers with WS who had
195 similar general cognitive profiles further supported the presence of intra-individual
196 differences in reading skills: one performed 8 years below chronological age on all word
197 reading, whereas the other was only 3 years below chronological age on word reading, and
198 was 2 years above chronological age on nonword reading (Dessalegn et al., 2013). Some
199 studies have reported nonword reading skills comparable to those of controls matched for
200 reading age and receptive vocabulary (Laing et al., 2001) or mental age (Garayzábal & Cuetos,
201 2008), whereas other studies have found lower nonword reading skills than those of mental
202 age-matched controls (Menghini et al., 2004). Reading skills (word identification) can
203 therefore be considered as relatively preserved in some individuals with WS, but impaired in
204 others. A consensus has nonetheless been reached on the existence of a lack of automatization
205 of reading processes (Barca et al., 2010; Levy et al., 2003; Steele et al., 2013).

206 *1.3. Predictors of reading ability in Williams syndrome*

207 The variability in reading skills raises the question of the specificity of the
208 neuropsychological profile of WS. For those individuals who can read, it is important to
209 understand the factors underlying their reading skills. Measuring an individual's reading
210 comprehension does not make it possible to identify problems emerging in the course of
211 reading acquisition (e.g., difficulties with the lexicon, word identification, implementation of
212 high-level processing or working memory). It is therefore essential to start by checking
213 whether word identification and the underlying skills are correctly acquired.

214 Reading acquisition requires the coordination and interaction of a number of cognitive
215 processes and mechanisms, such as oral language, phonological awareness, auditory-verbal
216 memory, and both auditory and visual perception (see Frith's cognitive model; Frith, 2001).
217 For instance, to master the alphabetic code, prereaders need to segment phonemes explicitly
218 into spoken words and recognize the previously learned graphic symbols that correspond to

219 the identified sounds. The metalinguistic skills of phonemic awareness (extracting the
220 relevant phonemic units from the continuous stream of speech heard) and visual analysis
221 skills are thus essential to reading acquisition (for a review, see Castles et al., 2018).
222 Mastering the decoding of grapheme-to-phoneme relationships enables printed words to be
223 translated into spoken language, thereby accessing information about meaning. If prereaders
224 have difficulty with symbol-sound mapping, reading acquisition will be limited to
225 memorizing the meanings of printed words, and it is unlikely that this strategy can be scaled
226 up to a full vocabulary (for a review, see Marinus & Castles, 2015). Building an orthographic
227 lexicon is therefore fundamental for lightening readers' mental load, enabling them to
228 establish a higher level of processing.

229 In WS, three hypotheses have been put forward to explain reading variability: one
230 general assumption relating to intellectual functioning (Garayzábal & Cuetos, 2008; Howlin
231 et al., 1998; Levy et al., 2003), and two specific hypotheses regarding phonological and
232 visuospatial skills (Brawn et al., 2018; Dessalegn et al., 2013; Garayzábal & Cuetos, 2008;
233 Levy et al., 2003). It should be noted that these hypotheses are not incompatible: deficits in
234 phonological and visual skills may exacerbate reading deficits attributable to an intellectual
235 deficit.

236 *1.3.1. General intellectual functioning factors and reading skills*

237 Results indicate that a minimum threshold of reasoning skills is required in order to
238 access reading in WS (Levy et al., 2003; Mervis, 2009). For instance, Howlin et al. (1998)
239 reported that the nonreaders in their sample had a significantly lower full score IQ, verbal IQ
240 and perceptual IQ than the group who was able to read. More broadly, adults with WS who
241 have mild-to-moderate intellectual disability (IQ between 35 and 70) have the reading skills
242 of 7- to 9-year-old children (Garayzábal & Cuetos, 2008; Howlin et al., 1998). Levy et al.
243 (2003) found that individuals with an IQ above 70 read within the low-to-average range,

244 whereas those with a full-score IQ of 50-70 read at their IQ level. In individuals with a
245 severe-to-profound intellectual deficit ($IQ \leq 34$), Laing et al. (2001) found a reading level
246 equivalent to a verbal mental age of 4 years and 7 months. We can therefore conclude that in
247 individuals with WS, word identification (word and nonword reading) is significantly
248 correlated with IQ, suggesting that a low IQ can explain poor reading skills (Levy & Antebi,
249 2004). Nevertheless, a unitary view of overall intellectual functioning is not enough to
250 understand the reading variability in WS - or indeed in typical development (Connors, 2003).

251 *1.3.2. Phonological awareness factors and reading skills*

252 Phonological awareness of phonemes or syllables allows individuals to analyze and
253 manipulate sounds in words (by segmenting and blending sounds). These oral language skills
254 are considered to be the most predictive for word reading in French children with typical
255 development (Schuele & Boudreau, 2008). Specific decoding deficits or dyslexia can be
256 caused by deficits in phonological and/or auditory processing (Nithart et al., 2009). For
257 instance, poor readers have deficits in the retention of verbal material and use of phonological
258 codes (related to a lack of subvocal rehearsal in auditory-verbal memory; Dufva et al., 2001;
259 for a neuroimaging study, see Pugh et al., 2013). However, there is no definitive evidence that
260 phonological awareness skills precede reading acquisition (Castles, Wilson, & Coltheart,
261 2011). Moreover, authors recognize that some phonological awareness tasks may place too
262 many demands on metacognitive abilities in the context of intellectual disability (Cupples &
263 Iacono, 2002).

264 In WS, some studies have identified a relationship between reading skills and
265 phonological processing, including phonemic awareness and auditory-verbal memory (Brawn
266 et al., 2018; Laing et al., 2001; Levy et al., 2003; Majerus et al., 2003; Menghini et al., 2004;
267 Temple, 2003). Studies have reported a correlation between word reading and performance on
268 some phonological tasks in WS (syllable deletion, rime detection, phonemic awareness),

269 compared with groups matched on mental age (Laing et al., 2001; Menghini et al., 2004) or
270 reading age (Laing et al., 2001). Reading variability in WS therefore appears to be related to a
271 lack of phonological awareness. However, some weaknesses in phonological coding also
272 seem to stem from auditory-verbal memory deficits (Levy et al., 2003; Majerus et al., 2003;
273 Menghini et al., 2004; Temple, 2003). This variability can thus be explained by deficits in
274 phonological processes, in the area of metalinguistic and/or executive skills and, more
275 broadly, in the categorical perception of speech sounds (phonetic recognition) (Catterall et al.,
276 2006; Majerus et al., 2011). By contrast, phonological awareness did not seem to be a
277 significant longitudinal predictor of reading growth when early reading development was
278 explored in 26 children with WS aged 4-8 years (Steele et al., 2013).

279 *1.3.3. Visuospatial factor and reading skills*

280 Visual and perceptual skills are involved in the sequential processing of written words,
281 from identifying the letters, graphemes and whole words (size, general shape, orientation) to
282 coding their position in words or sentences (spatial relationships between letter strokes,
283 letters' position in the word) (Frey & Bosse, 2018; Frith, 2001; Habib, 2002; Sprenger-
284 Charolles & Ziegler, 2019). Furthermore, a visual attention window is activated during the
285 analytical (letter, syllable or grapheme) or global processing of the written word (for
286 computational models, see Ans et al., 1998; Bosse et al., 2007). More broadly, the
287 identification of orthographic irregularities also depends on visual analysis skills (Bosse et al.,
288 2007; Castles et al., 2018). Neuroimaging studies featuring a written exposure task have
289 confirmed the involvement of visual skills, by pointing out the activation of the visual word
290 form area (VWFA) in the occipitotemporal ventral visual area (Dehaene et al., 2010).
291 Knowledge about letters, including perceptual invariance, sensitivity to letter combinations
292 and orthographic patterns, indeed appears to be clustered in the VWFA (Dehaene et al.,
293 2010).

294 Studies report that visuospatial and executive deficits, such as deficits in visual
295 attention, are partly responsible for the reading disorders observed in some cases of
296 developmental dyslexia (Bosse et al., 2007; Lallier et al., 2010; Peyrin et al., 2011). In the
297 neurobiological model of developmental dyslexia (Ramus, 2004), reading disorders stem from
298 a left perisylvian brain abnormality that gives rise to phonological deficits affecting
299 phonological awareness, short-term verbal memory, and access to the phonological lexicon.
300 Through a *snowball effect*, these deficits can induce dysfunctions of the magnocellular system
301 that contribute to auditory and visual deficits and, subsequently, to disturbances in the
302 posterior parietal cortex, reinforcing the visual deficits. Phonological deficits can therefore
303 also induce auditory and visual specificities. This assumption seems to be supported by
304 neuroimaging studies showing that early deficits in phonemic representations (temporal
305 planum) cause atypical visual word form area development in children with dyslexia
306 (Monzalvo et al., 2012).

307 In WS, reading deficits may be related to deficits in the visual processing of words
308 (Brawn et al., 2018; Dessalegn et al., 2013; Levy et al., 2003). Dessalegn et al. (2013) found
309 that two 16-year-olds with WS (BMP and HFK) differed by more than five grade levels on
310 reading skills, despite having similar cognitive profiles (reasoning skills and phonological
311 awareness). HFK had equivalent performance on nonword and word reading, whereas BMP
312 performed better on nonword than word reading. Furthermore, BMP performed better on
313 letter processing, object orientations (perceiving or remembering), and ordering tasks than
314 HFK. The authors suggested that HFK's poor reading skills were coming from visuospatial
315 processing deficits (Dessalegn et al., 2013). Other researchers have suggested that reading
316 deficits are induced by deficits in eye saccades, such as poor saccadic control and shorter
317 adaptation saccades (Van Herwegen, 2015). Reading deficits in WS may therefore be
318 explained by weaknesses in visual and visuomotor skills (Dessalegn et al., 2013).

319 ***1.4. Objectives of the current study and hypotheses***

320 The present study was the first to explore the variability of reading skills, and especially
321 its predictors, among French-speaking children and adults with WS. Based on work in other
322 languages, we assumed that some factors are involved in reading development in WS.
323 Accordingly, several factors, rather than just one, probably contribute to the variability in
324 single-word identification (Levy et al., 2003). We assessed two major predictors of reading
325 skills: phonological awareness and visuospatial skills; and we also controlled for nonverbal
326 mental age (or nonverbal reasoning), reading skills (decoding and word recognition), and oral
327 language. The WS group was compared to two control groups matched on sex and either
328 nonverbal mental age (C_M) or reading level (C_R) (e.g. Laing et al., 2001; Menghini et al.,
329 2004).

330 Our first objective was to clarify the nature of similarities and/or differences in reading
331 skills, as well as phonological and visuospatial skills, between the WS group and the two
332 control groups (Dessalegn et al., 2013; Laing et al., 2001; Levy et al., 2003; Majerus et al.,
333 2003; Menghini et al., 2004; Van Herwegen, 2015). Our second objective was to explore how
334 phonological and visual differences are related to reading efficiency (word identification),
335 after controlling for chronological age, nonverbal mental age and vocabulary (receptive and
336 expressive). Variability in reading efficiency may or may not be solely dependent on either
337 phonological or visuospatial skills (e.g., Laing et al., 2001; Menghini et al., 2004; Temple,
338 2003). We tested for two predictions: the phonological and visuospatial performance of the
339 WS group would be similar to or higher than that of the C_M group and lower than that of the
340 C_R group (H1); and this performance would be related to reading skills in the WS group (H2).
341 Given the paucity of results regarding visuospatial skills in the literature, we expected
342 phonological awareness to be a better predictor of word identification than visuospatial skills
343 in the WS group, as well as in both control groups. We also explored whether the links

344 between phonological or visuospatial performance and reading skills were similar for the WS
345 group and for matched controls.

346

347 **2. Method**

348 **2.1. Participants**

349 We recruited 28 French-speaking individuals with WS (14 females and 15 males; mean
350 age = 15.70 years, $SD = 8.10$, range = 7.07-36.10). All participants had been diagnosed with
351 WS based on genetic (fluorescence in situ hybridization (FISH) method), clinical and physical
352 examinations (facial dysmorphology, cardiovascular and musculoskeletal system anomalies).
353 As WS is rare, samples are traditionally small and the age distribution wide (e.g., Laing et al.,
354 2001; Menghini et al., 2004; Steele et al., 2013). We recruited our participants with WS via
355 the Autour des Williams association, Williams France federation, and genetic centers.

356 We used the Assessment of Cognitive Functions and Learning (Billard & Touzin,
357 2012) to control for a number of parameters that may be involved in reading profile
358 variability, beyond socio-economic and contextual factors: nonverbal reasoning (nonverbal
359 mental age), print exposure, vocabulary (receptive and expressive), reading skills (word
360 identification), and type of schooling. According to the parents' and participants' answers, all
361 individuals with WS had a medium socio-economic status, were (or had been) schooled in
362 France, and were monolingual. Just 10% received standard schooling with a special needs
363 teaching assistant (person helping them follow courses), while 45% divided their time
364 between traditional and specialized schooling, 14% were in a specialized educational
365 structure, and 31% were in a specialized vocational structure. Most participants with WS
366 performed below the 5th percentile on Raven's colored progressive matrices (using norms for
367 the closest age group), and would thus be considered as having an intellectual disability. Two

368 participants performed at the 10th percentile, two performed around the 50th percentile, and
369 one performed around the 75th percentile.

370 To examine differences in the involvement of phonological and visuospatial skills in
371 word identification (H1 & H2), participants with WS were matched with control groups on
372 sex and either nonverbal mental age (C_M) or reading level (C_R). Mental age was measured
373 using Raven's Colored Progressive Matrices, and reading level was measured using the
374 frequent word (FW) reading and PW reading subtests of the Assessment Battery of Written
375 Language (described in Section 2.2). Participants with WS were matched with C_M on
376 performance ± 1 point on Raven's matrices, and with C_R participants on performance ± 3
377 points on both frequent word reading and pseudoword reading.

378 A total of 518 control participants were tested for this study, and those who matched a
379 participant with WS were kept in the final sample. The C_M group comprised 90 typically
380 developing children (42 girls and 48 boys; mean age = 5.41 years, $SD = 1.17$, range =
381 3.06-8.04). The C_R group comprised 102 typically developing children (42 girls and 60 boys;
382 mean age = 6.84 years, $SD = 1.30$, range = 5.07-11.09 years). All participants with WS had at
383 least one matched control in both the C_M group and the C_R group; the median number of
384 controls was three per participant with WS per control group. In other words, the median
385 participant with WS had six matched controls in total.

386 All controls were typically developing French-speaking children, recruited from
387 schools in the French city of Rennes. They had the same socio-economic and contextual
388 characteristics as the WS group. All control participants were in mainstream schooling and
389 none had repeated a grade. Controls whose mental age differed from their chronological age
390 by more than 12 months were excluded (this is one of the screening criteria for intellectual
391 disability in France; INSERM, 2016).

416 respectively reflect the word recognition and decoding routes. The words' main
417 characteristics were length (short: 5 letters; long: 9-12 letters) and orthographic complexity
418 (complex words contained more letters per syllable than simple ones). For each lexical
419 category (FW/PW), we used lists of words containing short and simple (e.g., /image/ or
420 /fumal/), short and complex (e.g., /brune/ or /piète/), long and simple (e.g., /littérature/ or
421 /panacillane/), or long and complex (e.g., /merveilleux/ or /obyptienne/) words. In the FW
422 lists, 14 were nouns (6 concrete, 8 abstract, and 1 gender inflection), six were adjectives (all
423 abstract with no inflection), and four were verbs (3 concrete, 1 abstract with no inflection).
424 Participants had to read the words aloud as quickly as possible. We counted the number of
425 words that were correctly read (/24 points for each category).

426 2.2.2. *Covariates*

427 2.2.2.1. *Mental age (nonverbal reasoning)*

428 We used Raven's Colored Progressive Matrices test (Raven et al., 1998) to match
429 participants with controls on nonverbal mental age in the C_M group. This test was
430 standardized on a typical French population aged 4-11.5 years (for its psychometric qualities,
431 see Matthews, 1988). It comprises three sequences (A, Ab and B) of 12 items of increasing
432 complexity: Sequence A is perceptual, Sequence Ab is figurative, and Sequence B is
433 conceptual. For each item, an incomplete matrix has to be completed by the participant with
434 one of six options. We calculated the number of correct answers (/36 points).

435 2.2.2.2. *Receptive and expressive vocabulary*

436 Receptive vocabulary was evaluated with the French adaptation of the Peabody Picture
437 Vocabulary Test-Revised (PPVT-R; Dunn et al., 1993; for its psychometric qualities, see
438 Bouchard & Fitzpatrick, 2009). Form A is composed of five training items and 170
439 experimental items of increasing difficulty. Participants are shown four black-and-white
440 images, and have to pick the one that corresponds to a French word read out by the

441 experimenter. The test stops when the participant fails on six out of eight consecutive items
442 (/170 points).

443 Expressive vocabulary was evaluated with the Picture Naming subtest of the Oral
444 Language, Written Language, Memory and Attentional Skills battery (L2MA2; Chevrie-
445 Muller et al., 2010; for its psychometric qualities, see Bouchard & Fitzpatrick, 2009).
446 Participants have to name images presented one by one by the experimenter. If a participant
447 does not respond within 4 seconds or responds incorrectly, the experimenter has to provide a
448 strictly phonetic cue (e.g., saying /j/ to evoke the word /joue/). This subtest is composed of 54
449 color images divided into four main categories: body parts (/7), geometric shapes (/7),
450 objects/animals (/30), and general (/10). Only the first three categories (/44 points) are used to
451 gauge oral production skills.

452 *2.2.3. Determinants of reading acquisition: phonological and visuospatial skills*

453 *2.2.3.1. Phonological awareness*

454 Phonological awareness was evaluated with seven subtests from the L2MA2 (Chevrie-
455 Muller et al., 2010; for its psychometric qualities, see Bouchard & Fitzpatrick, 2009). Syllable
456 awareness was tested with two subtests: syllable segmentation (pronouncing all the syllables
457 in a PW; e.g., /bilu/), and deletion (deleting the first or last syllable of a PW; e.g., /kinu/).
458 Phonemic awareness was measured with five subtests: initial and median phoneme
459 identification (identifying the first or middle sound of a PW; e.g., /poub/ or /panr/), phoneme
460 segmentation (identifying all the sounds of a PW; e.g., /pal/), and initial and median phoneme
461 substitution (replacing the first or middle letter of a PW; e.g., /vour/ or /kat/). We counted the
462 numbers of correct answers for the syllable (/10 points) and phonemic (/35 points) awareness
463 subtests.

464 2.2.3.2. *Visuospatial abilities*

465 We used the Geometric Figure Copy subtest (GFC) of the L2MA2 (Chevrie-Muller et
466 al., 2010; for its psychometric qualities, see Bouchard & Fitzpatrick, 2009) to assess
467 visuoconstructive skills. Participants had to reproduce 15 geometric figures. We counted the
468 number of correctly reproduced figures (/46 points: the first 14 on 3 points, and the last one
469 on 4 points).

470 2.3. *Data Analysis*

471 To analyze differences in mean performance between groups (H1), we tested the effect
472 of group as a categorical independent variable on performance. If the effect was significant
473 when we simultaneously analyzed the three groups, we performed two follow-up analyses
474 comparing the WS and C_M groups and the WS and C_R groups. To look for differential
475 relations between reading skills and their predictors (phonological awareness and visuospatial
476 abilities) in the three groups (H2), we tested the interaction between group (categorical
477 variable) and each predictor of reading skills (continuous variable), which enabled us to see
478 whether the effect of predictors of reading skills varied significantly as a function of group.

479 Inspection of descriptive statistics revealed that phonological awareness for syllables
480 and phonemes, as well as for FW reading and PW reading, had non-normally distributed
481 residuals. These variables were rank-transformed prior to data analysis (all results were
482 comparable when retaining the non-transformed variables; inspection of the residuals
483 confirmed that rank-transforming the variables substantially improved normality). For the test
484 of mean differences between groups (H1), FW reading and PW reading were considered
485 separately. For the test of predictors of reading skills (H2), because these two variables were
486 very highly correlated ($r = .88-.97$ in the three groups), they were averaged to form a single

487 *reading skills* composite variable which was then rank-transformed (all results were
488 comparable when these two variables were analyzed separately).

489 Statistical analyses were performed using mixed linear modeling: this is equivalent to
490 classic regressions and analyses of variances, except that the analysis takes clustering of
491 participants into account. In the context of our design, where each participant with WS was
492 matched to multiple controls, this was the only solution that avoided violating the assumption
493 of independence between all participants. This was achieved by defining 28 clusters of
494 participants, with each cluster comprising one participant with WS and their matched
495 controls. This resulted in clusters of variable size, with the median cluster for a given
496 participant with WS including three controls matched on nonverbal mental age and three
497 controls matched on reading level. All analyses then included a random intercept specific to
498 each cluster of participants, thus accounting for non-independence between the matched
499 participants. All analyses also included group (WS, C_M or C_R) as a fixed effect. Models with
500 and without the parameter of interest were compared using chi-square tests. All analyses were
501 performed using the lme4 package for R (Bates et al., 2015).

502

503 **3. Results**

504

505 Descriptive statistics for mean performance in the three groups are displayed in
506 Table 2. A first series of analyses examined significant differences between groups for each
507 variable, by comparing a model including group as a fixed effect to a model not including
508 group. The results are displayed in Table 3. Overall, participants with WS were significantly
509 older than both C_M and C_R. Their nonverbal reasoning performance was significantly lower
510 than that of the C_R group, whereas their reading skills were significantly higher than those of
511 the C_M group, for both FW and PW reading. The receptive vocabulary of participants with
512 WS was significantly higher than that of the C_M group, but comparable to that of the C_R
513 group. Conversely, their expressive vocabulary was comparable to that of the C_M group, but
514 significantly below that of the C_R group.

515

516 [Insert Table 2 approximately here]

517

518 Our first hypothesis predicted that participants with WS would exhibit specificities in
519 terms of the predictors of reading skills considered here, namely phonological awareness for
520 phonemes and syllables, and visuospatial ability. The statistical tests summarized in Table 3
521 showed that this was indeed the case. Participants with WS scored significantly higher than
522 the C_M group for phonological awareness of both phonemes and syllables. They scored
523 significantly below the C_R group for phonological awareness of phonemes, but had similar
524 performance for phonological awareness of syllables. As for visuospatial ability, participants
525 with WS were comparable to the C_M group, but scored significantly lower than the C_R group.

526

527 [Insert Table 3 approximately here]

528

529 Our second hypothesis predicted that phonological awareness and visuospatial ability
530 would be related to reading skills in the WS group, and possibly to a greater extent than in the
531 C_M and C_R groups. We first explored the relations between the various measures and reading
532 skills using bivariate correlations, which are summarized in Table 4. Consistent with our
533 hypothesis, phonological awareness for both phonemes and syllables, as well as visuospatial
534 ability, were all related to reading performance in the WS group. This was also the case for
535 the two groups of matched controls. As for the other measures, chronological age predicted
536 reading skills in the two control groups, but not in the WS group. This was expected, given
537 these participants' lifelong learning of reading. Nonverbal reasoning was strongly predictive
538 of reading skills in all three groups. Receptive and expressive vocabulary scores were poorer
539 predictors of reading skills in the WS group than in the two control groups. Receptive
540 vocabulary in particular was a good predictor of reading skills in the C_R group, but not in the
541 WS group.

542 [Insert Table 4 approximately here]

543 Bivariate correlations suffer from two major issues in this context: they do not account
544 for potential covariates such as age, reasoning ability or vocabulary, which could inflate
545 correlations between variables; and they do not test whether the relation between variables is
546 significantly different across groups. To further explore these results, we thus tested the
547 contributions of vocabulary to reading skills when controlling for chronological age and
548 reasoning ability as covariates, and the contributions of phonological awareness and
549 visuospatial ability to reading skills when controlling for chronological age, reasoning ability,
550 and vocabulary as covariates. We also tested whether these contributions differed as a
551 function of group by testing whether there was a significant interaction between these
552 predictors and the group variable, indicating significantly different slopes between groups. In
553 other words, the following analyses tested the effect of possible predictors on reading skills in

554 models including predictors, group, the interaction between the two, and select covariates that
555 could artificially inflate effects, using mixed linear models.

556 The results of these analyses are summarized in Table 5. Phonological awareness for
557 phonemes was the single best predictor of reading skills in the three groups when controlling
558 for other variables. There were no significant differences between groups. Phonological
559 awareness for syllables was not a significant predictor for either the WS group or the C_R
560 group; it was however a significant predictor in the C_M group. A marginally significant
561 interaction with group confirmed that phonological awareness was a marginally better
562 predictor in the C_M group. A similar pattern emerged for visuospatial ability: it was not a
563 significant predictor for either the WS or the C_R group, but it was a significant predictor in the
564 C_M group, and the interaction with group was significant. Vocabulary was a poor predictor of
565 reading skills in all three groups when controlling for age and reasoning ability. There was a
566 significant difference between groups for receptive vocabulary, driven by a somewhat higher
567 correlation in the two control groups, but all correlations were nonsignificant.

568

569 [Insert Table 5 approximately here]

570

571 **4. Discussion and perspectives**

572 There is considerable heterogeneity between readers and prereaders with WS. To explain
573 this variability, it is important to study word identification skills before examining
574 comprehension skills. Phonological and visuospatial contributions were also considered
575 simultaneously in the study of Dessalegn et al. (2013) among English-speaking participants
576 with WS, but no study had previously been conducted in French, another language with a
577 deep orthographic system. The present study was the first to explore variability in single-word

578 reading among French-speaking children and adults with WS, and to compare the results with
579 those of two control groups matched for nonverbal mental age and reading level (under the
580 reasoning that comparisons with reading level rather than mental age may be more relevant
581 when studying the factors contributing to reading variability; e.g., Karmiloff-Smith et al.,
582 1997). By controlling for a number of developmental variables (chronological age, nonverbal
583 mental age, receptive and expressive lexicon) we were able to pinpoint some of the factors
584 responsible for differences in the mastery of reading skills for single-word identification by
585 individuals with WS. In this discussion, we focus on four aspects of the results: patterns of
586 reading skills in individuals with WS; the role of phonological skills; the role of spatial skills;
587 and the role of vocabulary.

588

589 *4.1. Single-word identification skills in WS*

590 Our results show that French-speaking individuals with WS are able to learn to read with
591 the appropriate teaching. From a cross-linguistic perspective, some differences became
592 clearer. Unlike young Spanish and Italian speakers who showed deficits in word recognition
593 (e.g., Garayzábal & Cuetos, 2008) and decoding (e.g., Barca et al., 2010; Menghini et al.,
594 2004), respectively, our sample's French speakers with WS exhibited word identification
595 skills (word recognition and decoding skills) appropriate for their nonverbal mental age,
596 despite French being an opaque orthographic system.

597 To clarify the characterization of reading procedures in French WS speakers, we
598 observed the lexicality, length and complexity effects. First, in the WS and C_R groups, reading
599 performance was lower for PW than for FW, in contrast to the C_M group (see Table 3). This
600 points to a relatively typical development of single-word identification skills in French
601 speakers with WS, as PW reading is typically more difficult than FW reading, owing to the
602 gradual implementation of the self-teaching mechanism (e.g., Castles et al., 2018; Share,

603 2004; Sprenger-Charolles & Ziegler, 2019). This observation means that there is a lexicality
604 effect among individuals with WS, who found FW easier to read than PW, as did the C_R
605 group. Second, qualitative results indicated greater difficulty reading long and complex words
606 than short and simple words. This reflected the presence of length and complexity effects in
607 the WS group, as in the C_R group. These results show the ongoing automatization of decoding
608 in WS, as well as in typically developing children matched on reading level (e.g., Barca et al.,
609 2010; Levy et al., 2003; Steele et al., 2013).

610 Overall, the level of single-word reading in the WS group was well above their level of
611 reasoning skills and single-word recognition appeared to be a relative strength despite a lower
612 nonverbal mental age than the C_R group (difference of 6.56 points or 2 years). We noted
613 differences in PW reading between the WS and C_R groups, with the WS group performed
614 worse than the C_R group. The use of two matching criteria to constitute the C_R group, namely
615 FW and PW reading, may explain this difference (see Table 3), as the groups were matched
616 on performance \pm 3 points on both FW and PW reading, possibly contributing to the 2-points
617 difference on PW reading.

618 Consistent with the literature, we also found a positive correlational link between single-
619 word identification and nonverbal reasoning in the WS group, as in both control groups (see
620 Table 4) (e.g., Brawn, et al., 2018; Howlin et al., 1998; Levy et al., 2003; Mervis, 2009).
621 However, no effect of chronological age was observed in the WS group, unlike the two
622 control groups. Although the reading skills of individuals with WS improve with age and
623 schooling, they do not correspond to their chronological age and their print exposure (e.g.,
624 Conners, 2003; Laing et al., 2001). These findings suggest that individuals with WS exhibit
625 delayed rather than atypical development of single-word identification.

626 To better understand the factors involved in the reading accuracy in the WS group, we
627 examined four types of reading errors: (1) no semantic errors (semantic relationship between
628 target and response), but (2) visual errors (responses shared the first letter or 50% of letters
629 with the target) in the form of substitutions (e.g., ‘stire’→‘strie’) or omissions (e.g., ‘tonil’→
630 ‘toni’), (3) visuossemantic errors resulting mainly in a change of the final phoneme (e.g.,
631 ‘gentillesse’→‘gentil’), and (4) decoding errors (e.g., ‘image’→‘imague’) (for reading errors,
632 see Dessalegn et al., 2013; Temple, 2003). All errors were made at an equal rate during PW
633 reading, whereas only visuossemantic and decoding errors were made during FW reading in
634 the WS group. These qualitative observations point to the implication of phonological factors
635 (decoding errors) and visual factors (visual and visuossemantic errors) during single-word
636 recognition, despite executive factors (visuomotor and/or phono-articulatory coordination).

637

638 Overall, proficient single-word identification in French-speaking individuals with WS
639 may be more consistent with the results of English case studies suggesting a reading profile
640 similar to that of profound dyslexia, including greater reading deficits for FW vs. PW (Levy et
641 al., 2003; Temple, 2003). Further studies focusing more specifically on word identification in
642 French-speaking individuals with WS are needed. Our first qualitative observations of reading
643 errors in the WS group confirm the involvement of phonological and visual factors, and
644 probably executive factors. Deficits of these factors may explain lower reading skills in WS.

645

646 ***4.2. Phonological contribution to learning to read in WS***

647 Phonemic awareness was the best predictor of single-word reading in the WS group after
648 controlling for developmental determinants (chronological age, nonverbal reasoning,
649 vocabulary), as was also the case in the C_R group (see Table 5). It was also an excellent
650 predictor in the C_M group, though visuospatial skills were slightly more predictive. In contrast

651 to the English-language longitudinal study by Steele et al. (2013), who found that phonemic
652 awareness did not appear to be a significant longitudinal predictor of reading growth in WS,
653 our results indicated that typical and atypical French-speaking readers share a similar profile
654 with phonemic awareness being predictive of reading skills (e.g., Castles et al., 2011; for
655 research on dyslexia, see Nithart et al., 2009; Ramus, 2004). The differences between these
656 studies may be contingent on the phonological task used by the authors (Steele's study tests
657 epi-phonology rather than metaphonology), but also on the variability in the phonological
658 factors involved in learning to read depending on the depth of the reference orthographic
659 system, even for two opaque languages (English and French). In French typical development,
660 phonemic awareness is fundamental to understanding the alphabetic principle, which is the
661 first step toward automatizing decoding (e.g., Sprenger-Charolles & Ziegler, 2019). During
662 development, phonological deficits may be reinforced by reading disorders, in addition to
663 being considered as risk factors (e.g., for typical development, see Castles et al., 2011; for
664 dyslexia, see Nithart et al., 2009; Ramus, 2004).

665 Our results seem to be consistent with the notion of typical but delayed development of
666 reading skills in the WS group, possibly related to early weaknesses in the alphabetic
667 principle implementation. These weaknesses may be induced by phonemic awareness
668 deficits (i.e., phoneme identification, segmentation and substitution performance) in the WS
669 group compared to C_R group, (see Table 3) (see, for example, Brawn et al., 2018; Garayzábal
670 & Cuetos, 2008; Levy et al., 2003; Menghini et al., 2004). By contrast, syllable awareness
671 appeared to be a strength in the WS group (e.g., Garayzábal & Cuetos, 2008; Menghini et al.,
672 2004; Rhodes et al., 2011). In addition, reading skills were positively correlated with
673 phonemic awareness (see Tables 4 and 5). Thus, phonemic awareness deficits seem to be both
674 the cause and the consequence of reading disability in the WS group.

675 Deficits in phonetic contrast perception of speech sounds (allophonic perception) can
676 lead to weak phonemic discrimination and, therefore, exacerbate these phonological deficits
677 (phonemic awareness) (e.g., Catterall et al., 2006; Majerus et al., 2011). Incorrect detection or
678 use of phonemic information therefore results in incomplete and unstructured phonological
679 representations in individuals with WS (e.g., Garayzabal & Cuetos, 2008; Karmiloff-Smith et
680 al., 1997; Majerus, 2019). In line with the neurobiological model (Ramus, 2004), the literature
681 on WS reports general deficits in phonological processing, such as auditory-verbal memory
682 (e.g., articulatory loop deficits; see Majerus et al., 2003; Menghini et al., 2004) and slow
683 lexical retrieval (Monnery et al., 2002). All these phonological deficits (phonemic awareness,
684 auditory-verbal memory and lexical retrieval) prevent phonological unit manipulation,
685 phonological coding and the construction of stable phonological representations in individuals
686 with WS.

687 Our results regarding receptive vocabulary (preserved in the WS group) and expressive
688 vocabulary (altered in the WS group; see Table 3) are respectively consistent with a
689 preservation of phonological and phono-semantic representations, and a deficit of phono-
690 articulatory coordination (e.g., Mervis, 2009; Monnery et al., 2002; Ypsilanti et al., 2005).
691 Thus, we suggest that in the WS group there may be executive deficits involved in the
692 slowdown of lexical retrieval (especially phonological), and not in the phonological
693 representation's alterations, as is also the case in certain profiles of dyslexia (e.g., Ramus,
694 2004) and in dysphasia (e.g., Lahey & Edwards 1999). This is also broadly consistent with
695 studies indicating delayed lexicosemantic development in WS, but no alteration in lexico-
696 semantic representations (e.g., Bello et al., 2004; Martens et al., 2008; Mervis, 2009).

697 The phonological processing specificities (phonemic awareness and auditory-verbal
698 memory deficits, and slow lexical retrieval) of individuals with WS, associated with larger
699 executive deficits, may therefore hinder the implementation of decoding, and thus prevent the

700 self-teaching of reading (e.g., on dyslexia see Ramus, 2004). At a higher level, phonological
701 deficits may reflect atypical development in the perceptual processing of phonological units in
702 WS. Longitudinal studies should be conducted to investigate the links between phonological
703 skills and perceptual processing. For example, phonological deficits are the main cause of a
704 specific reading disorder (dyslexia), although they can lead to perceptual processing (auditory
705 and visual) impairments that are secondarily involved in other reading disorders (e.g.,
706 neurobiological model; see Ramus, 2004).

707

708 *4.3. Visuospatial contribution to learning to read in WS*

709 Our measure of visuospatial and especially visuo-constructive skills correlated with
710 reading skills in all three groups (e.g. Brawn et al., 2018), but did not predict any variance in
711 single-word reading in the WS and C_R groups after controlling for developmental
712 determinants (chronological age, nonverbal reasoning, vocabulary), contrary to the C_M group
713 (see Table 5). Based on the control groups results, we suggest that during development the
714 visuospatial factors involvement in single-words reading could change from a cause /
715 consequence link to a consequence only link. In WS, altered visuospatial skills may be the
716 consequence rather than the cause of reading disability, in line with research on dyslexia (e.g.,
717 Ramus, 2004). As with some dyslexia profiles, participants with WS may have specificities in
718 the visual processing of written words (e.g., for dyslexia, see Bosse et al., 2007; Lallier et al.,
719 2010; Peyrin et al., 2011). According to the literature, copying performance is lower for
720 children with WS than for typical 5-year-olds (e.g., Heiz, 2019). We found that visuospatial
721 performance in the WS group was poorer than that of the C_R group, but similar to that of the
722 C_M group (e.g., Heiz & Barisnikov, 2016; Martens et al., 2008). The evidence points to
723 delayed visuospatial development in WS, rather than atypical development (e.g., Heiz &
724 Barisnikov, 2016; Martens et al., 2008).

725 The precise nature of the visuospatial deficits (deficit of visuospatial representation,
726 and/or deficit of general processes such as planning and motor programming) has yet to be
727 ascertained (e.g., Heiz & Barisnikov, 2016). In our study, we have assessed visuoconstructive
728 abilities in WS, but not elementary visual abilities. Regarding graphomotor skills, qualitative
729 analyses of GFC performance in the WS group revealed 1) thick, discontinuous, and
730 imprecise lines indicating poor motor coordination (e.g., Wuang & Tsai, 2017), 2) difficulty
731 with graphic production (proportions, orientations, locations, intersections between lines,
732 parallel and perpendicular lines, details) (e.g., D'Souza et al., 2016; Heiz & Barisnikov, 2016;
733 Rondan et al., 2008), and 3) difficulty copying complex shapes (integration of two or more
734 simple shapes), but not simple ones (lines, crosses, squares and triangles), in contrast to both
735 control groups (e.g., Heiz & Barisnikov, 2016). The poor GFC performance of the WS group
736 may thus be explained by graphomotor deficits, impaired perceptual integration (poor
737 comprehension of spatial relations between different pattern elements), as well as by deficits
738 in both early perceptual processing (processing of patterns' elementary characteristics: size,
739 orientation or slope) and intermediate-to-late perceptual processing (simultaneous local and
740 global processing) (e.g., Heiz, 2019). Deficits in all these dimensions of visuo-constructive
741 skills - motor, visuoperceptual and perceptual integration - may therefore be the consequence
742 of the reading disorder in WS, with the defect in visual strategy caused by the reading
743 disorder leading to impaired development of perceptual systems (e.g., for dyslexia, see Habib,
744 2002).

745 In typical development, elementary visual and visuoperceptual skills are necessary to
746 recover visual linguistic elements (size, general shape and orientation of graphemes) and their
747 visuospatial relationships, allowing for the subsequent identification and coding of their
748 position in a word or a sentence through visuo-attentional processing (e.g., Ans et al., 1998;
749 Frey & Bosse, 2018; Frith, 2001; Sprenger-Charolles & Ziegler, 2019). However, visuo-

750 attentional processing skills are recognized to be a *visual* cause of reading disorder (e.g.,
751 Bosse et al., 2007). This suggests that visuoperceptual and perceptual integration specificities
752 in WS may reduce the visuo-attentional window during reading and, consequently, hamper
753 the processes and mechanisms of letter and word identification (e.g., for typical development,
754 see Frey & Bosse, 2018). This hypothesis can be related to research highlighting a deficit in
755 the strategies of alternating between global and local processing of visuospatial information in
756 WS (Rondan et al., 2008). These visuospatial processing peculiarities may result from a deficit
757 in attentional disengagement in WS, at the same time giving weight to the hypothesis of the
758 *cascade effect of executive deficits* developed by Hoffman et al. (2003).

759

760 Phonemic awareness therefore seems to be the best predictor of single-word reading in
761 the present study, but elementary visual factors and attentional engagement need to be
762 explored further in WS. As in some forms of dyslexia, cerebral specificities (left perisylvian
763 abnormalities) can lead to phonological deficits in WS, which may give rise to other brain
764 dysfunctions, inducing auditory and visuoperceptual deficits (e.g., for WS, see Van
765 Herwegen, 2015; for dyslexia, see Monzalvo et al., 2012). This assumption will need to be
766 tested in future studies exploring early perceptual processing and neural networks during
767 reading in WS. Furthermore, mental rotation deficits in complex perceptual situations or ones
768 that concern subtle differences between two elements may induces imprecise orthographic
769 representations (e.g., Farran et al., 2003). Again, longitudinal studies are needed to examine
770 the development of orthographic representations in individuals with WS.

771

772 ***4.4. Lexical contribution to learning to read in WS***

773 Despite support for the hypothesis that expressive vocabulary deficits are a consequence
774 of the learning disability rather than a cause, the issue of the role of vocabulary remains

775 unresolved in WS (e.g., Martens et al., 2008; Monnery et al., 2002; Ypsilanti et al., 2005; for
776 research on dyslexia, see Ramus, 2004). Our results showed no effect of vocabulary
777 (receptive and expressive) on the variability of single-word identification in the WS group,
778 and no effect in the two control groups (see Table 5) when controlling for select covariates.
779 The reading level of participants with WS seemed to be lower than their receptive vocabulary
780 level (9.2 years), compared with both the C_R group (8.10 years) and the C_M group (8.1 years)
781 (see Table 3) (e.g., Barca et al., 2010; Laing et al., 2001). Learning to read appeared to have
782 an effect on increasing vocabulary level in the two control groups; there is a consequential
783 link between reading and lexical level.

784 We hypothesize that individuals with WS exhibit rapid planning deficits between the
785 phonological form of the chosen word in long-term memory and its articulation (see section
786 4.2). These specificities may induce a lexical retrieval which is much too slow during reading.
787 The literature supports this hypothesis, by showing that the reduced lexical access in WS
788 hinders the linguistic development (morphology, syntax, semantic and pragmatic) that
789 determines access to reading comprehension regardless of orthographic system depth:
790 transparent in Italian (Bello et al., 2004; Burani et al., 2006; Vicari et al., 2004) and opaque in
791 English (Mervis, 2009; Perovic & Wexler, 2007; Ring & Clahsen, 2005; Stojanovik, 2006)
792 and French (Monnery et al., 2002). Thus, as the single-word reading tasks are timed, it is
793 possible that participants with WS did not take the time to rely on their lexical knowledge
794 during their reading; this type of task is very time-consuming for them. Longitudinal studies
795 are needed to better understand the nature of lexical determinants involved in learning to read
796 in WS.

797

798 **5. Conclusion**

799 The unique neuropsychological profile of people with WS may provide a better
800 understanding of the causes and consequences of reading disorders: it is possible that both
801 phonological and visuospatial weaknesses, associated with larger executive deficits, hinder
802 the implementation of decoding, and thus prevent the self-teaching of reading in WS. Our
803 study among French-speaking readers, confirms that phonemic awareness is the best predictor
804 of single-word identification. Variability in single-word identification in the WS group may
805 thus be explained by the interaction between the various phonological deficits (phonemic
806 awareness, auditory-verbal memory and lexical access). We suggest that learning to read
807 exacerbates early phonological deficits in WS, or at least makes them more salient.
808 Visuospatial deficits do not appear to contribute to variability of reading skills within the WS
809 group when controlling for other variables, and may be a consequence of the reading disorder
810 rather than their cause.

811 Our study controlled for some developmental variables (chronological age, nonverbal
812 reasoning, vocabulary), but these results need to be reinforced by longitudinal studies, in
813 order to clarify the developmental trajectories of the factors that contribute to written
814 language acquisition. Although it is difficult to control some dimensions, including the extent
815 of print exposure or age span, results show that it is better to compare the performance of
816 individuals with WS with that of controls matched on reading level rather than either
817 chronological age or nonverbal mental age. This method can also be applied to many other
818 populations with intellectual disabilities. Despite their atypical intellectual functioning,
819 French-speaking individuals with WS are able to learn to read with the appropriate teaching.
820 A better understanding of phonological and visual risk factors, as well as the impact of a
821 reading disorder on these factors, would enable us to tailor literacy instruction to individual

822 specificities. Reading remediation must become widespread to improve functioning in many
823 aspects of individuals' everyday lives.

824

825

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828

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ANNEXES

1059 **Table 1.** Print exposure (years) and number of participants (*n*) for each of the three groups
1060 (WS, C_M, C_R).

	WS (<i>n</i> = 29)	C _M group (<i>n</i> = 90)	C _R group (<i>n</i> = 102)
Number of participants for each print exposure duration	10-18 years, <i>n</i> = 13 7-9 years, <i>n</i> = 6 5 years, <i>n</i> = 2 4 years, <i>n</i> = 5 3 years, <i>n</i> = 3	2 years, <i>n</i> = 13 1 year, <i>n</i> = 21 1 year of awareness, <i>n</i> = 19 0 years, <i>n</i> = 37	4-6 years, <i>n</i> = 6 2-3 years, <i>n</i> = 38 1 year, <i>n</i> = 38 1 year of awareness, <i>n</i> = 20

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1063 **Table 2.** Descriptive statistics for all measures for the three groups (WS, C_M, C_R)

	WS group (<i>n</i> = 28)			C _M group (<i>n</i> = 89)			C _R group (<i>n</i> = 87)		
	<i>M</i>	<i>SD</i>	Range	<i>M</i>	<i>SD</i>	Range	<i>M</i>	<i>SD</i>	Range
Chronological age	15.70	8.10	7 - 36	5.41	1.17	3 - 8	6.84	1.30	5 - 11
Nonverbal reasoning	18.29	5.70	9 - 32	18.45	4.98	9 - 32	24.85	4.94	15 - 33
Frequent word reading	14.82	7.39	1 - 24	5.42	8.74	0 - 24	15.87	7.91	1 - 24
Pseudoword reading	9.75	6.02	1 - 21	4.20	7.02	0 - 22	11.75	6.40	1 - 21
Receptive vocabulary	100.75	24.40	43 - 140	90.34	24.68	31 - 148	97.98	20.50	44 - 142
Expressive vocabulary	36.11	4.83	21 - 43	37.70	8.69	13 - 53	42.67	7.20	23 - 60
Phonological awareness - phonemes	19.32	9.95	0 - 35	10.35	13.23	0 - 35	25.17	9.73	2 - 35
Phonological awareness - syllables	9.25	0.93	7 - 10	7.08	3.00	0 - 10	9.18	1.23	3 - 10
Visuospatial ability	13.18	9.05	1 - 38	14.84	7.53	0 - 33	24.90	8.94	9 - 44

1064 *Note.* Chronological age is expressed in years. Nonverbal reasoning is the raw score on
 1065 Raven's CPM (0-36). Pseudoword and frequent word reading are raw scores on the BELEC
 1066 (0-24). Receptive vocabulary is the raw score on the PVVT-R (mean 100, standard deviation
 1067 15). Expressive vocabulary is the raw score on L2MA2 Picture Naming (0-44). Phonological
 1068 awareness for syllables and phonological awareness for phonemes are raw scores on the
 1069 L2MA2 (0-10 and 0-35). Visuospatial ability is the raw score on the GFC (0-46).

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1072 **Table 3.** Mean differences between groups

	WS vs. C _M vs. C _R	WS vs. C _M	WS vs. C _R
Chronological age	$\chi^2(2) = 156.26, p < .001$	$\chi^2(1) = 92.55, p < .001$	$\chi^2(1) = 71.85, p < .001$
Nonverbal reasoning	$\chi^2(2) = 78.07, p < .001$	$\chi^2(1) = 2.88, p = .090$	$\chi^2(1) = 32.44, p < .001$
Frequent word reading	$\chi^2(2) = 93.51, p < .001$	$\chi^2(1) = 31.27, p < .001$	$\chi^2(1) = 0.13, p = .719$
Pseudoword reading	$\chi^2(2) = 68.26, p < .001$	$\chi^2(1) = 17.58, p < .001$	$\chi^2(1) = 6.33, p = .012$
Receptive vocabulary	$\chi^2(2) = 7.34, p = .025$	$\chi^2(1) = 3.82, p = .050$	$\chi^2(1) = 0.79, p = .373$
Expressive vocabulary	$\chi^2(2) = 22.16, p < .001$	$\chi^2(1) = 1.15, p = .284$	$\chi^2(1) = 19.02, p < .001$
Phonological awareness - phonemes	$\chi^2(2) = 69.84, p < .001$	$\chi^2(1) = 13.15, p < .001$	$\chi^2(1) = 11.20, p < .001$
Phonological awareness - syllables	$\chi^2(2) = 48.37, p < .001$	$\chi^2(1) = 18.53, p < .001$	$\chi^2(1) = 0.09, p = .766$
Visuospatial ability	$\chi^2(2) = 74.43, p < .001$	$\chi^2(1) = 1.61, p = .205$	$\chi^2(1) = 39.84, p < .001$

1073 *Note.* This table represents the results of comparisons between a model including group:
 1074 dependent variable $\sim 1 + \text{Group} + (1 | \text{Cluster})$; and a model not including group: dependent
 1075 variable $\sim 1 + (1 | \text{Cluster})$.

1076

1077 **Table 4.** Bivariate correlations between reading skills and their predictors for the three groups

	WS group ($n = 28$)	C _M group ($n = 89$)	C _R group ($n = 87$)
Chronological age	-.07	.68 ***	.69 ***
Nonverbal reasoning	.59 ***	.53 ***	.44 ***
Receptive vocabulary	.30	.23 *	.60 ***
Expressive vocabulary	.28	.47 ***	.39 ***
Phonological awareness - phonemes	.72 ***	.65 ***	.65 ***
Phonological awareness - syllables	.39 *	.57 ***	.38 ***
Visuospatial ability	.60 *	.62 ***	.44 ***

1078 *Note.* * $p < .05$. ** $p < .01$. *** $p < .001$.

1079

1080 **Table 5.** Differential relations between reading skills and their predictors for the three groups

	WS β	C _M β	C _R β	Predictor*Group interaction test statistic	<i>Predictor*Group interaction p-value</i>
Receptive vocabulary	-.21	.15	.09	$\chi^2(2) = 6.82$	$p = .033$
Expressive vocabulary	-.12	.19	.06	$\chi^2(2) = 3.29$	$p = .190$
Phonological awareness - phonemes	.52	.45	.35	$\chi^2(2) = 1.19$	$p = .550$
Phonological awareness - syllables	.18	.34	.12	$\chi^2(2) = 4.94$	$p = .084$
Visuospatial ability	.10	.54	.06	$\chi^2(2) = 13.20$	$p = .001$

1081 *Note.* This table represents the results of analyses predicting reading skills, using models of
 1082 the form: reading skills ~ 1+Group+Predictor+Group:Predictor+Covariates+(1|Cluster). The
 1083 covariates included chronological age and nonverbal reasoning in all analyses, as well as
 1084 receptive and expressive vocabulary when testing for the effects of phonological awareness
 1085 and visuospatial ability. β coefficients indicate the standardized slope for the effect of a
 1086 predictor on reading skills in a given group; significant slopes are in bold. The test statistic
 1087 and p value indicate whether this slope differed as a function of group.