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Predicting reading and spelling disorders: a 4-year prospective cohort study

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7 **Keywords:** reading disorder, spelling disorder, predictors³, phonological awareness⁴, invented
8 spelling⁵, textual competence⁶.

9 Abstract

10 In this 4-year prospective cohort study, children with a reading and spelling disorder, children with a
11 spelling impairment, and children without a reading and/or spelling disorder (control group) in a
12 transparent orthography were identified in third grade, and their emergent literacy performances in
13 kindergarten compared retrospectively. 642 Italian children participated. This cohort was followed
14 from the last year of kindergarten to third grade. In kindergarten, the children were assessed in
15 phonological awareness, conceptual knowledge of writing systems and textual competence. In third
16 grade, 18 children with a reading and spelling impairment and 13 children with a spelling impairment
17 were identified. Overall, conceptual knowledge of the writing system was the only statistically
18 significant predictor of the clinical samples. No differences were found between the two clinical
19 samples.

20

21 1 Introduction

22 Spelling disorders have often been found to be associated with reading disorders (Lyon et al., 2003),
23 a finding that is further supported by the consideration that reading and spelling performances are
24 also associated in the general population (Bates et al., 2006). The existence of associations between
25 disorders poses questions about whether they share the same cognitive basis (Pennington, 2006).
26 Furthermore, studies on reading and spelling disorders need to take the level of consistency of the
27 mapping between letters and sounds in words into account as a level of explanation, and increase our
28 understanding of transparent orthographies (Ziegler et al., 2010). This 4-year prospective cohort
29 study compared, in kindergarten, the early cognitive skills of a sample of spelling-disabled pupils
30 (SD) with those of a sample of reading-and-spelling-disabled pupils (RSD) and with those of a
31 sample of children without a reading and/or spelling disorder (control group). The study was
32 conducted in an Italian-speaking population and is characterised by the fact that Italian provides a
33 transparent orthography. A better knowledge of the differences in the early cognitive skills between

34 these three groups of children can contribute to identifying the predictors of spelling impairments,
35 which are still underspecified and poorly understood (American Psychology Association, 2013).

36 **1.1 Definition of reading and spelling disorders**

37 In line with the findings suggesting an association between learning disorders — e.g. between
38 reading (dyslexia) and spelling disorders (dysorthographia) (Egan and Tainturier, 2011; Lyon et al.,
39 2003; Moll et al., 2014) — the latest edition of the American Psychiatric Association’s Diagnostic
40 and Statistical Manual of Mental Disorders (DSM-5) combines the DSM-IV diagnoses of a number
41 of disorders: reading disorder, mathematics disorder, disorder of written expression and learning
42 disorder not otherwise specified (American Psychology Association, 2013). The DSM-5, however,
43 stresses the possibility of a dissociation between these different learning disorders (Berninger et al.,
44 2015), as it requires separate coding of deficits belonging to specific domains. Thus, dyslexia is
45 defined as a learning disorder that produces an impairment in reading and requires the specification
46 of whether word reading accuracy, reading rate or fluency, spelling, or reading comprehension are
47 compromised (ICD-9 code: 315.00; ICD-10 code: F81.0). Likewise, dysorthographia is defined as a
48 learning disorder with an impairment in written expression, and it requires the specification of
49 whether spelling accuracy, grammar and punctuation accuracy, clarity, or organization of written
50 expression are compromised (ICD-9 code: 315.2; ICD-10 code: F81.81). Following the indication of
51 the DSM-5 (American Psychology Association, 2013), in this study we identified two clinical
52 groups: (1) children with a specific learning disorder with an impairment in reading accuracy and
53 fluency (315.00), which was associated with a specific learning disorder with an impairment in
54 written expression, in particular in spelling accuracy (315.2), and (2) children with a specific learning
55 disorder with an impairment in written expression only, in particular in spelling accuracy (315.2).
56 These disorders were diagnosed in absence of comorbidity with other neuro-developmental (e.g.,
57 ADHD) or mental disorders (e.g., anxiety disorder) that typically co-occur with specific learning
58 disorders.

59 **1.2 Spelling in reading and writing**

60 Interestingly, the term “spelling” is used for both reading and writing. Whereas the use of spelling
61 disorder for a writing disorder is quite obvious, many influential definitions of the reading disorder
62 also include spelling problems in children (Lyon et al., 2003; Pennington, 2009), as well as in adults
63 (Afonso et al., 2015). For example, according to the International Dyslexia Association and National
64 Institutes of Child Health and Human Development, a reading disorder is characterized by difficulties
65 with accurate and/or fluent word recognition and by poor spelling and decoding abilities.

66 By focusing on the spelling impairment, this study's overall aim is to contribute to a better
67 understanding of the association between reading and spelling disorders. In fact, spelling is a
68 bridging skill between reading and writing which, if impaired, produces a reading-writing disorder.
69 However, spelling is asymmetrical, as it is more difficult when writing than when reading. Thus, a
70 mild spelling impairment may allow pupils to master the easier process (*i.e.*, reading), but not the
71 more difficult one (*i.e.*, writing). Conversely, a severe spelling impairment may cause pupils to
72 struggle in both processes, reading and writing. According to past research, a specific writing
73 impairment might be a residual problem of those pupils who have managed to compensate for earlier
74 reading difficulties (Newman et al., 1993). Studies on spelling disorders vs. reading-spelling
75 disorders are lacking, mostly because research on reading disorder has focused on reading only, thus
76 neglecting its relation with spelling disorders (Morken and Helland, 2013).

77 **1.3 The role of the transparency of the writing system**

78 Reading and spelling disorders change depending on the level of transparency of a writing system
79 (i.e. how consistently letters map onto sounds —Paulesu et al., 2001; Raman and Weekes, 2005). In
80 transparent writing systems (e.g. Italian or German), in which each letter is almost always
81 pronounced in the same way in different words, the typical problem of children with a reading
82 disorder is reading fluently, rather than accuracy (Zoccolotti et al., 2014; 2015; Barca et al., 2006).
83 Conversely, in opaque writing systems (e.g. English or French), in which some letters are
84 pronounced in different ways in different words, children with a reading disorder struggle to read
85 fluently and also correctly (Wimmer and Mayringer, 2002; Wimmer and Schurz, 2010). Instead,
86 children with a spelling impairment are inaccurate writers in both orthography systems, transparent
87 and opaque (Angelelli et al., 2010). It should also be noted that, in most languages, spelling is more
88 difficult than reading (Newman et al., 1993). This difficulty gap is enhanced in transparent
89 orthographies, in which the regularity of the orthographic system is higher in grapheme-phoneme
90 relations (forward regularity) than in phoneme-grapheme relations (backward regularity)
91 (Notarnicola et al., 2012; Wimmer and Mayringer, 2002) — for example, in Italian the phoneme /k/
92 can correspond to two different graphemes, ‘c’ as in /'kwoko/ (‘cuoco,’ en. tr. ‘chef’), or ‘q’ as in
93 /kwi/ (‘qui,’ en. tr. ‘here’).

94 The Italian language, because of its characteristics of transparency and reading-spelling asymmetry,
95 provides optimal conditions to study spelling impairment as an independent disorder, and spelling
96 impairment in association with a reading impairment. In addition, Italian spelling in writing plays a
97 leading role for the acquisition of both, reading and writing (Pinto et al., 2015), which makes the
98 exploration of the early predictors of this process even more crucial.

99 **1.4 Predictors of reading and spelling disorders**

100 In this study, children with a spelling disorder (SD), children with a reading and spelling disorder
101 (RSD), and children without a reading/spelling disorder (control group) were identified in third
102 grade. Their emergent literacy performances in the last year of kindergarten were then retrospectively
103 compared. According to Pennington (2006), in fact, finding a common antecedent deficit would
104 confirm the severity hypothesis, according to which RSD is an earlier and more severe form of the
105 same etiology underlying the SD. In this paragraph, we discuss the literature on the predictors of
106 reading and spelling disorders.

107 Although spelling has not received a similar amount of research interest as reading, there are several
108 studies available on predictors of spelling, also in transparent orthographies. Many of these studies
109 support the existence of different cognitive predictors of reading and spelling. According to Vaessen
110 and Blomert (2013), among the most important predictors of reading, only phonological awareness
111 (i.e. the ability to identify and manipulate units of sounds) and letter-sound matching skills (i.e. the
112 ability to match letters to corresponding speech sounds) are also predictors of spelling, especially in
113 transparent orthographies.

114 Among the aforementioned skills, phonological awareness is the most debated, in particular
115 concerning its relationship with the acquisition of reading and spelling skills across different
116 languages. For quite some time, phonological awareness had been considered to be the most
117 important predictor of reading (Paulesu et al., 2001) and spelling acquisition (Babayiğit and
118 Stainthorp, 2007; Vaessen and Blomert, 2013). Recently, however, several researchers have
119 questioned its status in transparent orthographies, in both normal acquisition of reading and spelling

120 (Babayiğit and Stainthorp, 2007) on the one side, and in learning disorders (Bigozzi et al., 2016;
121 Wimmer and Schurz, 2010) on the other one. A better understanding of the role of phonological
122 awareness in reading and writing thus requires the assessment of phonological awareness before the
123 onset of formal literacy, since conventional acquisition of reading and writing exerts an
124 autoregressive effect on phonological awareness (Nikolopoulos et al., 2006).

125 Letter-sound matching skills are particularly important for reading fluency in beginner readers
126 (Vaessen and Blomert, 2013), but fluency quickly reaches full development in transparent
127 orthographies, which reduces the importance of letter-sound matching skills. In opaque (Caravolas et
128 al., 2001) and transparent orthographies (Landerl and Wimmer, 2008; Torppa et al., 2013), instead,
129 letter-sound matching skills remain associated to later spelling performances, although the effect-size
130 of this association has been questioned, on the basis of the argument that knowing which letter
131 belongs to which speech sound is not as important as using this knowledge efficiently and
132 automatically (Vaessen and Blomert, 2013). Finally, in contrast with the clear association between
133 RAN and reading disorders in transparent orthographies (Torppa et al., 2013), the theoretical link
134 between RAN and spelling is also debated (Babayiğit and Stainthorp, 2007; Nikolopoulos et al.,
135 2006; Torppa et al., 2013; Vaessen and Blomert, 2013).

136 Interest in the beginning stages of literacy development has focused attention on the very early
137 invented spelling created by young children prior to formal reading and spelling instruction. Invented
138 spellings, meant both as children's early attempts at writing (Read, 1971) and as children's early
139 attempts at reading (Liberman, 1971), have been considered as a marker of children's phonological
140 awareness, and of their knowledge of the phonemic segments (sounds) represented by an alphabet.
141 This assumed that since pre-reading children did not have a visual image of words fixed in their
142 memory, when they sought to represent words they did so based on articulatory features.

143 Several authors have claimed that literacy outcomes are better predicted by an association between
144 phonological awareness and letter knowledge, rather than by tasks tapping into oral phonological
145 skills only (Pinto et al., 2009; Hulme and Snowling, 2013; Ouellette and Sénéchal, 2008; Wimmer
146 and Schurz, 2010). Blaiklock (2004) contributed to the understanding of the combination of
147 phonological-orthographic representations in kindergarten by demonstrating that the orthographic
148 representations of words actually mediate the relationship between phonological awareness and
149 literacy processes. Pinto et al. (2009) also suggested that children's conceptual knowledge of the
150 writing system captures this interplay between phonological and orthographic representations of the
151 words, strongly predicting literacy acquisition.

152 Typically, conceptual knowledge of the writing system is assessed by an invented spelling task, in
153 which the participant creates sound-signs that correspond to their level of knowledge of the writing
154 system, from simple signs that discriminate writing from drawing, to an awareness that longer words
155 require more signs than shorter words, to a 1:1 correspondence between sounds and signs in a word,
156 although signs are not alphabetically correct. This early cognitive skill refers to phonological-
157 orthographic connectivity and encompasses the systematic (even if not conventional) matching of
158 sounds with written letters, and the productive component of writing, the ability to graphically build
159 and develop a stable pattern of orthographic signs (even if unconventional and incorrect). In this
160 sense, this factor takes into account the combined contribution of phonological awareness with other
161 skills that are related to literacy acquisition and impaired in children with a reading disorder, that is
162 grapho-motor skills (see Berninger et al., 2008), and visual attention (see Germano et al., 2014).

163 Conceptual knowledge of the writing system includes child's knowledge of the print conventions, of
164 the names of letters, and of the letter sounds (Niessen et al., 2011).

165 Notwithstanding recent advances in research on conceptual knowledge of the writing system, its
166 unique contribution to children's acquisition of reading and spelling needs to be better understood
167 (Niessen et al., 2011). Our research on emergent literacy predictors of reading and reading disorders
168 (Bigozzi et al., 2016), and spelling (Pinto et al., 2009) in the Italian language has found that, when
169 the conceptual knowledge of a writing system was included with phonological awareness among
170 kindergarten predictors, the predictive power of phonological awareness disappeared, probably
171 because its effect was absorbed by the conceptual knowledge of the writing system and integrated
172 with orthographic knowledge. These results bring further evidence to Wimmer and Schurz's
173 hypothesis that reading disorders are better explained by an early deficit in orthographic-
174 phonological connectivity (2010). Conceptual knowledge of the writing system is also a better
175 predictor of reading and reading disorders (Bigozzi et al., 2016), and spelling in writing (Pinto et al.,
176 2009), than children's textual competence. In an emergent literacy perspective, textual competence is
177 an ability that is inter-related with other kindergarten competences, and is considered a
178 developmental precursor to conventional forms of reading and writing (Lonigan et al., 2000). Thus,
179 the ability to connect the phonological and orthographic representations of a word, (*i.e.*, conceptual
180 knowledge of the writing system) seems to be a more important cognitive skill for predicting reading
181 and writing acquisition than the ability to get to grips with the individual units of meaning conveyed
182 by the word and to form a network of relations between words that are in the text (*i.e.*, textual
183 competence).

184 **1.5 Aims of the study**

185 The aim of this study was to determine whether RSD and SD shared the same predictive pattern in
186 kindergarten in terms of emergent literacy skills. In particular, (1) we focused on children's
187 conceptual knowledge of the writing system, and (2) we tested in a transparent writing system
188 whether the conceptual knowledge of the writing system is an antecedent of RSD and SD children's
189 common impairment in spelling, similarly to what was found for reading acquisition and reading
190 disorders (Bigozzi et al., 2016). We also studied the role of phonological awareness, because its
191 predictive role for reading and spelling skills in transparent orthographies is debated.

192 The Italian language, which is a transparent writing system, allows to explore the relationship
193 between emergent literacy and reading and spelling disorders, and fill the gap with our understanding
194 of such a relationship in the context of opaque languages (e.g., English). In addition, the higher
195 degree of transparency in the sign-sound correspondence in comparison with the sound-sign
196 correspondence, allows one to clearly identify two clinical groups, RSD and SP, and run a
197 comparative analysis between them and with the reference population.

198 The present study addressed these aims by carrying out a 4-year prospective cohort study. From a
199 methodological perspective, a prospective cohort study shares the advantages of a longitudinal
200 approach. However, previous longitudinal studies on reading and spelling disorders included only
201 pupils from the population at risk of SD or RSD (e.g. familiarity or specific language impairment, see
202 for instance Lyytinen et al., 2004), but excluded all those children with reading and/or spelling
203 disorders that are present in the population not at risk. We designed a prospective cohort study so as
204 to include all children from the natural population, at-risk and not-at-risk for learning disorders. From
205 this general population, the SD and RSD samples were extracted from the same cohort, and were
206 compared to the same control group. This approach provides a better control of potentially

207 confounding variables (e.g. socio-economic status), and allows to better understand the relation
208 between reading and spelling disorder. A prospective cohort study presents a further advantage. It
209 allows to assess predictors of reading and spelling disorder symptoms manifesting in the 3rd grade
210 among children's early skills in kindergarten, before the onset of formal literacy (*i.e.*, before
211 children's early skills are influenced by the autoregressive effect of conventional learning of reading
212 and spelling in primary school).

213 We expect the RSD and SD groups to show an impaired conceptual knowledge of the writing system
214 in kindergarten, when compared to the control group (hypothesis 1). We expect the RSD and SD
215 groups to show no impairment in phonological awareness or textual competence, when compared to
216 the control group (hypothesis 2). Finally, we expect the SD and RSD groups to show no significant
217 differences between each other in phonological awareness, conceptual knowledge of the writing
218 system, and textual competence (hypothesis 3).

219 **2 Material and Methods**

220 **2.1 Participants**

221 We followed a cohort of 642 Italian children from a mid-sized city in Central Italy (mean age:
222 $4.98 \pm .31$ years; 299 girls and 343 boys) for 4 years, from the last year of kindergarten to the third
223 grade. From this sample, we had previously excluded children showing a formal mastery of reading
224 and writing during kindergarten. The parents of the participants gave informed consent for the
225 participation of their children in the study. The measures were administered at a time agreed upon
226 with the school and with due adherence to the requirements of privacy and informed consent required
227 by the Italian law (Law Decree DL-196/2003). Regarding the ethical standards for research, the study
228 referred to the last version of the Declaration of Helsinki (World Medical Association, 2013). The
229 present study was approved by the Ethical Committee of the Department of Psychology at the
230 University of Firenze, Italy. In the third grade, from the cohort of children, three groups were
231 identified: 18 RSD pupils (12 boys and 6 girls), 13 SD pupils (9 boys and 4 girls), and 611 normally-
232 reading and -spelling pupils (322 boys and 289 girls). Interestingly, the two clinical samples
233 respected the boy:girl ratio typically found in the literature for both reading and spelling disorder
234 (Moll et al., 2014). Thus, the control group (children without a reading and/or spelling disorder) also
235 presented a prevalence of boys over girls.

236 In the Italian educational system, children typically start kindergarten at the age of three, and finish it
237 when they are five. Children then start primary school when they are six years old. Primary school
238 lasts five grades. The school year begins in mid-September and ends in mid-June. All classes
239 participating in the study (kindergarten and primary school) were part of the same school district
240 therefore they shared some characteristics: similar educational and teaching practices and middle
241 socio-economical level. Most importantly, in Italy the formal teaching of literacy begins in primary
242 school, and follows a specific curriculum, as set down in national law. All the participating
243 kindergartens were following the national guidelines issued by the Ministry of Education, which
244 were valid at the time of the study. Since all emergent literacy skills are strongly dependent on family
245 or kindergarten practices (Lonigan et al., 2000), we checked that no schools were following a specific
246 program on formal literacy, and that no participant was already able to read and write in a
247 conventional way at the time of the kindergarten assessment.

248 An important characteristic of Italian schools is low mobility: families tend to live in the same
249 neighborhood over several generations. Children generally attend school in the same area. Therefore,
250 in this study, subject attrition through the three stages was extremely low.

251 **2.2 Research design**

252 We present 4-year longitudinal data from a study of children from kindergarten to third grade.
253 Children's emergent literacy skills were assessed in kindergarten, at the beginning of the last school
254 year. Four years later, when the participants were in third grade, we singled out the pupils who had
255 received a diagnosis of reading and spelling disorder and the ones with a diagnosis of spelling
256 disorder, and retrospectively analyzed their emergent literacy skills, comparing their performances to
257 their normally-reading and normally-spelling peers. To ensure that all pupils had equal opportunity to
258 be flagged as RSD or SD, we checked that none of the children included in the control group had
259 received a diagnosis of a specific learning disorder.

260 The RSD and SD participants had received their diagnosis from the clinical units of the Italian
261 National Health System, which follows the International Classification of Mental Disorders, ICD-10
262 (World Health Organization, 1992). The clinical units gave the researchers of this study access to
263 each SD and RSD child's protocol, in accordance with local privacy laws and standards.

264 **2.2.1 Clinical groups**

265 In the following, we describe the criteria to be included in the SD or RSD group. Each SD and RSD
266 child had displayed difficulties learning and using academic skills for at least six months, despite the
267 provision of targeted interventions. SD displayed difficulties with written expression, with an
268 impairment in written spelling, grammar or punctuation, as assessed by the Battery for the
269 Assessment of Developmental Reading and Spelling Disorders (Sartori et al., 2007). RSD displayed
270 inaccurate and slow word reading, as assessed by MT Battery of Reading (Cornoldi and Colpo,
271 1998). With regard to cut-off scores, Moll et al. (2014) demonstrated that the association between
272 RSD and SD depends on what thresholds we set to decide who to include in the two clinical groups,
273 thus in this study we adopted strict criteria to form the groups. In the RSD group, children had a
274 reading accuracy and fluency score below the 5th percentile, as well as a written spelling
275 performance score below the 5th percentile. In the SD group children's writing accuracy was lower
276 than the 5th percentile, whereas their reading performance was above the 5th percentile (see table 1).
277 RSD and SD children did not show any intellectual disability, as assessed by the Wechsler
278 Intelligence Scale for Children-III (Wechsler, 2006), were not affected by uncorrected visual or
279 auditory acuity, any mental or neurological disorder, psychosocial adversity, lack of proficiency in
280 Italian or inadequate educational instruction. These aspects were assessed through the clinical
281 synthesis of the individual's history (developmental, medical, family and educational), school
282 reports, and psycho-educational assessment.

283 **2.2.2 Control group**

284 Children just failing to meet the cut-off points of pathological performance (e.g., a performance of
285 7th percentile) were kept in the control group as their reading and spelling was not impaired at a
286 clinical level, and represent a sample from the reference population. In Italy psychopathologies or
287 disabilities are identified by the local health authorities at the parents' request (Law 104/1992; Law
288 170/2010; Ministerial Decree 12 July 2011). After the diagnostic procedure ends, the local health
289 authority gives the papers to the parents, who deliver them to the school, so that the procedures of
290 school inclusion can be started (Decree of the President of the Council of Ministers 185/2006).

291 Specific learning disabilities can be detected by teachers too, by notifying the child's family so that
292 they can proceed to start a diagnostic procedure with the local health authorities (Inter-Ministerial
293 Ministry of Education, Universities and Research-Ministry of Health Decree; 17/4/2013)¹. At the
294 time of the study, control group children were not affected by any type of pathology, nor were they
295 included in a diagnostic procedure, or identified by the teachers as children with special educational
296 needs.

297 INSERT TABLE 1

298 **2.3 Measures**

299 Preschoolers were evaluated through tests measuring emergent literacy skills (phonological
300 awareness, textual competence and conceptual knowledge of the writing system). All the children's
301 products were coded by two independent judges. Agreement between the judges was between 88%
302 and 99%; cases of disagreement were resolved through discussion. All the measures reported
303 acceptable and good reliability scores.

304 **2.3.1 Phonological awareness**

305 Identification and production of sound patterns (Dowker and Pinto, 1993). The children were
306 exposed to two verbal stimuli, one containing rhymes, and the other a series of alliterating words.
307 The instruction was: "Now I am going to tell you a poem, which is a bit like a story but not quite.
308 And I would like you to make one up too." They were asked to produce a poem of their own, with the
309 stimuli acting as examples. The order of the two stimuli was counterbalanced. Three scores were
310 derived: rhythm (children's ability to reproduce the prosody); rhyme (children's ability to detect the
311 rhymes within the stimulus); and alliteration (children's ability to detect alliterations within the
312 stimulus). The alpha coefficient for this instrument was .82. From this test, three measures were
313 derived.

314 Identification and production of rhythm. The children's ability to reproduce the prosody (rhythm)
315 was scored as follows: 0 no rhythm produced, 1 one rhythm produced, 2 two or more rhythms
316 produced. Pupils' scores ranged from 0 to 2. Agreement between the judges was 94%.

317 Identification and production of rhyme. The children's ability to detect the rhymes within the
318 stimulus was scored as follows: 0 no rhymes produced, 1 one rhyme produced, 2 two or more rhymes
319 produced. Pupils' scores ranged from 0 to 2. Agreement between the judges was 97%. An example of
320 a poem with rhyme detection from a kindergarten participant was:

321 mi piacciono le farfalle [I like butterflies]

322 azzurre, rosse e gialle [blue, red, and yellow]

323 Identification and production of alliteration. The children's ability to detect alliterations within the
324 stimulus was scored as follows: 0 no alliterations produced, 1 one alliteration produced, 2 two or
325 more alliterations produced. Pupils' scores ranged from 0 to 2. Agreement between the judges was
326 98%. An example of a poem with alliteration detection from a kindergarten participant was:

¹ See <https://www.european-agency.org/country-information/italy/national-overview/identification-of-special-educational-needs> for more information on the identification of physical and mental disabilities and disorders in Italy

327 scivolano gli sciatori sciando [the skiers slide while they're skiing]

328 Identification of phonemes (Dowker and Pinto, 1993). The children were asked to identify similar
329 words among triplets of words, two of which had a phoneme in common. The alpha coefficient for
330 this instrument was .79. Agreement between the judges was 93%; cases of disagreement were
331 resolved through discussion. Children were exposed to nine three-word sets, and had to identify the
332 two words with the initial phoneme in common. In three series they had to identify the initial
333 phoneme (e.g. PALO - PESCA – NOTTE), in three series they had to identify the intermediate
334 phoneme (e.g. AGO - UGO – EVA), and in three series they had to identify the final phoneme (e.g.
335 BORSA - PRATO – TRENO). The following score was assigned: 0 if children correctly coded 0 to 2
336 triplets, 1 if children correctly coded 3 to 5 triplets, and 2 if children correctly coded 6 to 9 triplets.
337 Pupils' scores ranged from 0 to 2.

338 **2.3.2 Conceptual knowledge of a writing system**

339 Invented spelling (Pinto et al., 2009). The scoring procedure we developed aimed to measure the
340 extent to which an unconventional (e.g., incorrect) response made by a kindergarten child captured
341 two main features of the written alphabetic language: the phonetic structure of the words (i.e. the
342 number and the type of phonemes) that the child represented and the level of orthographic
343 representation he/she adopted., and were sensitive enough to classify the lower level responses of
344 kindergarten children. Children's early written productions were analyzed in a quantitative and also
345 qualitative manner using three categories, measuring the children's knowledge of the sound-sign
346 correspondence but also of the word boundaries, word morphology, directionality of print, number
347 and shapes of letters required/allowed to compose a word. The children were asked to draw and
348 write, from which three different scores were obtained. The alpha coefficient for this instrument was
349 .92. Two independent raters coded the children's products. The inter-rater reliability was 94%.
350 Disagreements were resolved by discussion between the two raters.

351 Conceptual knowledge of orthographic notation. The children were asked to write down their name,
352 the words they knew, and the word 'mela' (apple), for a minimum of two items. This score defined
353 how similar children's signs were to conventional letters. Scores were assigned as follows: 0 for
354 drawings, 1 for scribbles, 2 for forms similar to letters, 3 for sequences of well-shaped letters.

355 Conceptual knowledge of the orthographic variation of sound quantity. Children were asked to write
356 down two long words (one given by the experimenter, one of their choice), and two short words (one
357 given by the experimenter, one of their choice), for a total of four items. This score defined whether
358 the children were aware of the numeric correspondence between sounds and signs (one sign per
359 sound). Scores were assigned as follows: 0 for drawings; 1 for performances based on a non-
360 correspondence between signs and sounds (words of the same length, or the longer word written
361 shorter than the short word); 2 for performances in which the difference in length is present and
362 correct, without a 1:1 correspondence between signs and sounds; 3 for performances in which the
363 difference in length is present and correct, with a 1:1 correspondence between signs and sounds.

364 Conceptual knowledge of the orthographic variation of phonemic units. The children were asked to
365 write two pairs of words, each of which were formed by two words with the same first part and only
366 the last letter different, for a total of two items This score defined whether the children were aware
367 that words which sound similar are also written in a similar way, with small variations. Scores were
368 assigned as follows: 0 for drawings, 1 for performances in which the two words were written, either
369 identically, or completely differently; 2 for performances with a partial equivalence and a partial

370 differentiation, where the two parts do not correspond to sound variations, however; 3 for
371 performances with a partial equivalence and a partial differentiation, in which the two parts
372 correspond perfectly to variations in sounds.

373 2.3.3 Textual competence

374 Story production (Spinillo and Pinto, 1994). The children were asked to tell a narrative. In the Italian
375 school, kindergarten and primary school, this type of instruction refers to the production of fictional
376 stories. All participants understood the instructions well and produced fictional stories. The story was
377 recorded, transcribed and analyzed by two independent judges on three parameters: structure,
378 cohesion and coherence. The inter-rater reliability was 91%. Disagreements were resolved by
379 discussion between the two raters. The alpha coefficient for this instrument was .91.

380 Structure. The story structure was coded by eight elements: a) title, b) conventional story opening, c)
381 characters, setting, d) problem, e) central event, f) resolution, g) conventional story closing. The
382 system to attribute the structure scores was:

383 first level, non-story (1 point): simple descriptions of actions without any characteristics of narrative
384 style such as a conventional story opening or conclusion;

385 second level, sketch story (2 points): introduction of the setting and the main character, conventional
386 story opening is often present, but both the problem and resolution are missing;

387 third level, incomplete story (3 points): elementary narrative structure, setting and characters are
388 introduced, often with a conventional story opening and conclusion, but a central event is missing;

389 fourth level, essential story (4 points): non-essential structural elements, such as setting, are missing;

390 fifth level, complete story (5 points): all eight elements are included, with only the title considered
391 optional

392 Causal cohesion: to assess the causal cohesion in children's stories, all the causal linguistic elements
393 were identified (e.g. because, thus, so, and the like). On the basis of the quantity of causal cohesive
394 elements used in the stories, balanced by the total number of words, three increasing levels of causal
395 cohesion were identified: absent (0 points), low (1 point), medium (2 points) and high (3 points).

396 Temporal cohesion: to assess the temporal cohesion in children's stories, all the temporal linguistic
397 elements were identified (e.g. once upon a time, then, because, after that, therefore, and the like). On
398 the basis of the quantity of temporal cohesive elements used in the stories, balanced by the total
399 number of words, three increasing levels of temporal cohesion were identified: absent (0 points), low
400 (1 point), medium (2 points) and high (3 points).

401 Coherence: to analyze coherence in the children's narratives, the number of incoherencies were
402 identified. On the basis of the number of incoherencies, balanced by the total number of sentences,
403 three increasing levels of cohesion were identified: absent (0 points), low (1 point), medium (2
404 points) and high (3 points).

405 2.4 Data analysis

406 Each variable's extreme outliers were identified and eliminated by observing the relative box-plots.
407 Through examination of the skewness and kurtosis of each dependent variable's probability
408 distribution we verified that all variables were normally distributed. The statistical software R version
409 3.2.0 (R Core Team, 2015) was used to perform a linear mixed effects (LME) analysis of the
410 relationship between group type (SD, RSD or control group) and the notational knowledge of a
411 writing system, phonological awareness and textual awareness. Separate LME models were run for
412 each DV with the lmer function from the packages lme4 (Bates et al., 2014) and lmerTest
413 (Kuznetsova et al., 2015). Model fitting was done by employing restricted maximum likelihood
414 (REML). Compared to standard linear regression models, LME models are well suited for the
415 analysis of unbalanced data sets (e.g. Sikorska et al., 2015). LME analysis decomposes model effects
416 into the contribution of a fixed component (here the group) and a random component (here the class
417 nested within the school nested within the school district). By including random-effect factors, the
418 model can take the hierarchical structure linked to these factors into account.

419 Including a by-school within district and by-class within school within district random slope for the
420 group led to an overparameterized model (correlation of -1.00 or 1.00 of the intercepts and slopes for
421 the random effects), so we simplified the final models to include random intercepts for district, for
422 school within district, and for class within school within district, and by- district random slopes for
423 group. Collinearity was not an issue: all fixed-effect correlations ($|r|$) were less than .35.

424 The fixed effect estimates are provided by regression coefficients. To obtain an "effect size" of the
425 group effect on notational knowledge, phonological awareness and textual awareness, we computed
426 the LME standardized regression coefficients (β). When group membership is dummy coded with the
427 control group as the baseline, a change in group membership results in a change of β standard
428 deviations in the outcome. The standardized regression coefficients, therefore, provide a measure of
429 effect size akin to Cohen's d by taking the hierarchical nature of the data into account.

430 Visual inspection of residual plots did not reveal any obvious deviations from assumptions of
431 homoscedasticity or normality. p -values were obtained using the `pbkrtest` in R (Halekoh and
432 Højsgaard, 2014) for likelihood ratio test and parametric bootstrapping (with 10,000 resamples), and
433 the `multcomp` package (Hothorn et al., 2008) with a Tukey correction for multiple comparisons.

434 **3 Results**

435 **3.1 Descriptive results**

436 In table 2 pupils' performances (SD, RSD and control group) in kindergarten skills are reported.

437 INSERT TABLE 2

438 **3.2 Differences in predictors between SD, RSD and control group**

439 After applying a Box-Cox transformation to correct for skewness, a principal component analysis
440 (PCA) was performed on the centered and scaled variables describing the conceptual knowledge of a
441 writing system, that is, orthographic notation (FNScr), phonemic units (FNSuSe), and sound quantity
442 (FNVarNum). The first PC was used as an index of conceptual knowledge of the writing system
443 (CKWS 72% explained variance). The correlations between CKWS and FNScr, FNSuSe, and
444 FNVarNum were .86, .83, and .85, respectively. By using the same procedure, we created a
445 phonological awareness index (PA). The correlations between PA (74% of explained variance) and

446 the variables rhythm (CFRit), rhyme (CFRim), alliteration (CFAllPro) and phonemes (CFfon) were
 447 .92, .66, .94 and .89 respectively. Likewise, an index of textual competence (TC) was created. The
 448 correlations between TC (74% of explained variance) and the variables structure (StoStr), causal
 449 cohesion (StoCau), temporal cohesion (StoTem), and coherence (StoCoe) were .92, .66, .89 and .94,
 450 respectively. Table 3 reports the correlations between the three principal components, conceptual
 451 knowledge of the writing system, phonological awareness and textual competence.

452 INSERT TABLE 3

453 For conceptual knowledge of the writing system, including group in the model significantly increased
 454 the fit compared with a null, intercept-only model, $\chi^2_2 = 7.93$, $p = .0189$, $p(\text{bootstrap}) = .0204$,
 455 thus indicating a main effect of group. Tukey post hoc contrasts showed a statistically significant
 456 difference between the SD and control groups, $z = 3.39$, $p = .0023$, and between the RSD and control
 457 groups, $z = 2.73$, $p = .0166$, but not between the SD and RSD groups, $z = 1.63$, $p = .2268$. The β
 458 weights for the difference between the control group (baseline) and the RSD and SD groups were
 459 equal to -0.59 and -1.21 , respectively. Conditional R^2_{GLMM} (Johnson, 2014) was equal to .36
 460 (variance explained by both fixed and random factors), with 12% of the explained variance due to the
 461 fixed-effects factor (see Figure 1).

462 INSERT FIGURE 1

463 For phonological awareness, we found no main effect of group, $\chi^2_2 = 1.38$, $p = .5007$,
 464 $p(\text{bootstrap}) = .4510$; Conditional $R^2_{\text{GLMM}} = .21$, with .67% of the explained variance due to the
 465 fixed-effects factor (see Figure 2).

466 INSERT FIGURE 2

467 Likewise, we found no main effect of group for textual competence, $\chi^2_2 = .73$, $p = .6942$,
 468 $p(\text{bootstrap}) = .6471$; Conditional $R^2_{\text{GLMM}} = .21$, with 0.47% of the explained variance due to
 469 the fixed-effects factor (see Figure 3).

470 INSERT FIGURE 3

471 3.3 Analyses using matched control group

472 In a different set of analyses, we only selected control participants from the classes where either an
 473 SD or an RSD child was found, to control for the effect of relevant confounding variables, i.e. socio-
 474 economic status, educational environment and gender. Two separate control groups were created: one
 475 for SD children ($n = 62$) and one for RSD children ($n = 98$). When only one SD or RSD child was
 476 present in a class, or when SD or RSD children present in a class had the same gender, controls were
 477 also matched for gender.

478 LME models were used to examine the group difference between SD or RSD children as measured
 479 by the conceptual knowledge of the writing system, phonological awareness, or textual competence
 480 dependent variables, with the same random-effect structure as described before. SD children had
 481 lower conceptual knowledge of the writing system scores than school-matched controls, $\chi^2_1 = 10.37$,
 482 $p = .0019$; the β weights for the difference between the control (baseline) and the SD and RSD
 483 groups were equal to $-.46$ (s.e.=.22) and -0.87 (s.e.=.27), respectively. No statistically significant

484 difference was found between SD children and controls with respect to phonological awareness,
485 $\chi^2_{(1)}=.97, p=.3240$, or textual competence, $\chi^2_{(1)}=.92, p=.3385$.

486 RSD children also showed lower conceptual knowledge of the writing system scores than school-
487 matched controls, $\chi^2_{(1)}=.420, p=.0403$; no statistically significant difference was found between
488 RSD children and controls with respect to phonological awareness, $\chi^2_{(1)}=.12, p=.7272$, or textual
489 competence, $\chi^2_{(1)}=.49, p=.4824$.

490 **3.4 Reading performances in first grade**

491 To confirm the severity hypothesis, that specific spelling impairment might be a residual problem of
492 pupils who have compensated earlier reading difficulties, we examined the reading performances of
493 the three groups in first grade. According to the norms of the reading test used in this study (Cornoldi
494 and Colpo, 1998), the cut-off score to diagnose an impairment in reading fluency is .51
495 syllables/second (5th percentile). Control group pupils were reading .76 syllables/second ($\pm .12$). RSD
496 were already showing an impairment in reading in the first grade, as they were reading .40
497 syllables/second ($\pm .10$). Instead, SD pupils just failed to meet the cut-off score of pathological
498 performance ($.57 \pm .18$ syllables/second). In third grade, SD reading fluency performance improved
499 drastically (see table 1).

500 **4 Discussion**

501 This 4-year study followed a cohort of Italian children from the last year of kindergarten to the third
502 grade, when pupils were diagnosed with RSD or SD. Their kindergarten performance in conceptual
503 knowledge of the writing system, their phonological awareness, and their textual competence were
504 retrospectively compared to the performance of a control group peers. Our main findings are
505 described below.

506 **4.1 RSD and SD children versus NRS peers**

507 In kindergarten, SD and RSD children show an impaired conceptual knowledge of the writing system
508 relative to control children without a reading and/or spelling disorder. The results from this cohort of
509 children confirmed the results of a previous study on Italian children with a reading disorder (Bigozzi
510 et al., 2016), and extend those finding to SD pupils too. In two previous studies (Pinto et al., 2009;
511 2012) conceptual knowledge of the writing system was shown to be an important predictor of
512 spelling acquisition in first grade. This study extends the predictiveness of children's invented
513 spelling to the atypical learning trajectory of spelling too, as SD children were characterized by poor
514 performances in this measure. Moreover, we found no evidence of differences in phonological
515 awareness (in kindergarten) between SD, RSD and control group children, thus supporting the idea
516 that phonological awareness shows a limited power in predicting RSD (Bigozzi et al., 2016; Pinto et
517 al., 2015; Wimmer and Schurz, 2010). Our results thus suggest that SD and RSD are associated
518 disorders (Bates et al., 2006; Egan and Tainturier, 2011; Lyon et al., 2003).

519 That the conceptual knowledge of the writing system resulted to be the only statistically significant
520 predictor does not show that phonological awareness is unrelated to the development of spelling
521 skills (Babayiğit and Stainthorp, 2007; Vaessen and Blomert, 2013). Indeed, the conceptual
522 knowledge of the writing system is a complex task, which integrates different cognitive, perceptual
523 and grapho-motor activities, with a phonological load (phonological coding of the input,
524 identification of phonological units, ideation and choice of a transcoding system, and then execution

525 of the transcoding system). Thus, we speculate that phonological awareness is integrated within
526 conceptual knowledge of the writing system, rather than substituted by it, in agreement with previous
527 theories stating that this factor is the medium through which phonological awareness exerts its effect
528 on reading skills (Ouellette and Sénéchal, 2008). Given the multicomponential nature of conceptual
529 knowledge of the writing system, besides the phonological load, other components could contribute
530 to the predictivity of this factor on RSD and SD. For instance, the impairment could take place at the
531 level of the visual-motor integration (Adi-Japha and Freeman, 2001). Future studies should explore
532 these issues to increase our understanding of the specific contribution of conceptual knowledge of the
533 writing system.

534 **4.2 RSD children versus SD peers**

535 Our data show that SD and RSD children share a similar performance in phonological awareness and
536 textual competence, and similar impairment in conceptual knowledge of the writing system. This
537 result leaves still unanswered the question of whether the two clinical groups differ from each other
538 in performances in kindergarten predictors. SD and RSD pupils do not show any difference in terms
539 of performances in kindergarten skills.

540 We propose that RSD and SD children should be understood as belonging to two points on a
541 continuum, rather than having two distinct pathologies. Although RSD and SD have similar levels of
542 impairment in conceptual knowledge of the writing system, they show different spelling deficits,
543 with a different level of severity: a spelling disorder (low severity) and reading and spelling disorder
544 (high severity). We propose that this difference stems from the process of formal literacy. This
545 proposal is consistent with the idea that variations in reading and spelling performances are
546 influenced by many biological and contextual factors, (*e.g.*, literacy environment at home and quality
547 of instruction, see Hulme and Snowling, 2013).

548 The formalization and conventionalization that take place in primary school of the skills informally
549 involved in the conceptual knowledge of the writing system in kindergarten requires pupils to
550 perform two cognitive actions, spelling in writing and spelling in reading, with the former being
551 more difficult than the latter (Newman et al., 1993; Wimmer and Schurz, 2010). Because of the
552 asymmetry between the demands of spelling and reading in the formal setting, children diagnosed on
553 the basis of a specific reading impairment, typically have writing problems too, while other pupils
554 only have a significant impairment in writing. In this sense, we agree with Pennington's severity
555 hypothesis (2006) and Newman et al.'s (1993) residual problem hypothesis: the specific spelling
556 impairment might be a residual problem of pupils who have managed to compensate for earlier mild
557 reading difficulties. The analysis of participants' reading performances in first grade supports this
558 hypothesis, as SD pupils' reading performances just failed to meet the cut-off score of pathological
559 performance. However, the small sample sizes of the two clinical groups, SD and RSD, does not
560 allow us to exclude the existence of significantly differing levels of impairment in conceptual
561 knowledge of the writing system, which could also contribute to the potential explanation of the
562 differential manifestation of the spelling deficit in SD and RSD. These considerations might apply
563 specifically to transparent writing systems. If spelling and reading are asymmetric in all languages,
564 such asymmetry is enhanced in transparent writing systems (Notarnicola et al., 2012; Wimmer and
565 Mayringer, 2002). Indeed, opaque orthographies might induce higher rates of a combined reading
566 and spelling disorder, whereas transparent orthographies could create the conditions for children to
567 compensate their spelling difficulties when reading, especially by relying on the phonological route.

568 The main conclusion of this study is that RSD and SD children in a transparent writing system share
569 a common deficit among kindergartener's skills: conceptual knowledge of the writing system. As
570 Sampaio and Capellini (2014) highlighted, students who are exposed to literacy in a reflection-
571 focused way show better literacy performances, as the orthographic processes become automatic and
572 they can draw their attention to the content of the text, rather than to the correct spelling of it.
573 Longitudinal studies on later reading and spelling performances may help identify early cognitive
574 predictors, although it is important to note that such predictors do not determine disorders in an all or
575 nothing way, as developmental interactions among early cognitive skills are likely and concur with
576 the genetic risk of the manifestations of symptoms (Hulme and Snowling, 2013). At the practical
577 level, identifying a plausible cognitive variable predicting later literacy disorders is critical for
578 planning for educational intervention. Conceptual knowledge of the writing system could be a target
579 skill to be included in screening tools for early identification of reading and spelling disorders. To
580 this aim, future research should test its sensitivity (i.e., the proportion of true positives identified) and
581 specificity (i.e., the proportion of true negatives identified), to validate invented spelling as a
582 screening system (Andrade et al., 2015). An early intervention on skills that can potentially hinder
583 the acquisition of reading and spelling can decrease the possibility of negative outcomes, also
584 preventing a decrease in motivation and self-imposing restrictions on the literacy activities that
585 children with a learning disorder often exhibit.

586 This study had several limitations. We found that phonological awareness is predictive of RSD when
587 integrated with conceptual knowledge of the writing system. It would be interesting to consider
588 different measures for phonological awareness, besides those used in the present study, as the impact
589 of this construct on the prediction of reading and spelling disorders could depend on what component
590 is measured (see Germano and Capellini, 2011). Although we propose that reading and spelling
591 disorders share a common core (conceptual knowledge of the writing system), other explanations
592 may be possible, including impairment of other skills that are involved in the acquisition of
593 orthographic knowledge (e.g., RAN, such as the sensitivity to orthographic regularities, letter
594 knowledge and the implicit learning skills). Future studies should test the hypothesis that SD children
595 are pupils who had previous reading difficulties and managed to resolve them. Moreover, future
596 studies should also explore the factors contributing to help SD children to better cope with their
597 reading difficulties.

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- 745

746 **6 Tables**

747 Table 1

748 Cut-off scores, number and proportion of children falling below the cut-offs, reading speed
 749 (syllable/seconds), reading accuracy (number of errors), and writing accuracy (number of errors) of
 750 control group, RSD and SD children in third grade (mean, standard deviations and range)

	Cut-off (5 th percentile)	N (%)	Control group	RSD	SD
Reading speed	1.18	18 (2.80)	3.5±1.2 (1.55-5)	1.15±.50 (.90-1.18)	3±1.1 (1.50-3.90)
Reading errors	13	18 (2.80)	4.9±3.50 (0-6)	15±4.3 (13-19)	5±4.8 (1-6)
Writing errors	11	31 (4.83)	4.31±3.50 (0-8)	14.50±2.80 (13-18)	15.30±3.50 (14-20)

751

752 Table 2

753 Descriptive statistics of kindergarten measures: mean and standard deviation (minimum; maximum)

Construct	Measure	Control group	RSD	SD
Phonological Awareness	Rhythm	1.05±.76 (0;2)	1.38±.59 (0;2)	.92±.76 (0;2)
	Rhyme	1.13±.80 (0;2)	1.52±.60 (0;2)	1.23±.73 (0;2)
	Alliteration	.64±.75 (0;2)	.90±.63 (0;2)	.67±.78 (0;2)
	Phonemes	1.04±.76 (0;2)	1.19±.51 (0;2)	.92±.64 (0;2)

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	Notation	2.12±.65 (0;2.3)	1.50±.74 (0;3)	1.42±.73 (0;2.3)
Conceptual knowledge of a writing system	Sound quantity	1.54±.58 (0;2)	1.19±.66 (0;2)	1.17±.72 (0;2)
	Phonemic units	1.52±.91 (0;3)	1.14±.84 (0;3)	.88±.43 (0;1.5)
	Structure	2.04±1.53 (0;5)	1.71±1.23	1.67±.89 (0;3)
Textual competence	Causal cohesion	.76±.58 (0;3)	.86±.36 (0;1)	1.17±.58 (0;2)
	Temporal cohesion	1.28±.95 (0;3)	1.05±.67 (0;3)	1.00±.43 (0;2)
	Coherence	1.14±.69 (0;2)	1.00±.55	1.17±.72 (0;2)

754

755 Table 3

756 Correlations between the three principal components, conceptual knowledge of the writing system
757 (CKWS), phonological awareness (PA) and textual competence (TC).

	CKWS	PA	TC
CKWS	1.00		
PA	.26*	1.00	
TC	.23*	.28*	1.00

758 Note. *p<.0001

759

760

761

762 **7 Figures**

763 Figure 1. Plot representation of SD, RSD and control group in conceptual knowledge of the writing
764 system in kindergarten

765

766 Figure 2. Plot representation of SD, RSD and control group in phonological awareness in
767 kindergarten

768

769 Figure 3. Plot representation of SD, RSD and control group in textual competence in kindergarten

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Provisional

Figure 1.TIF

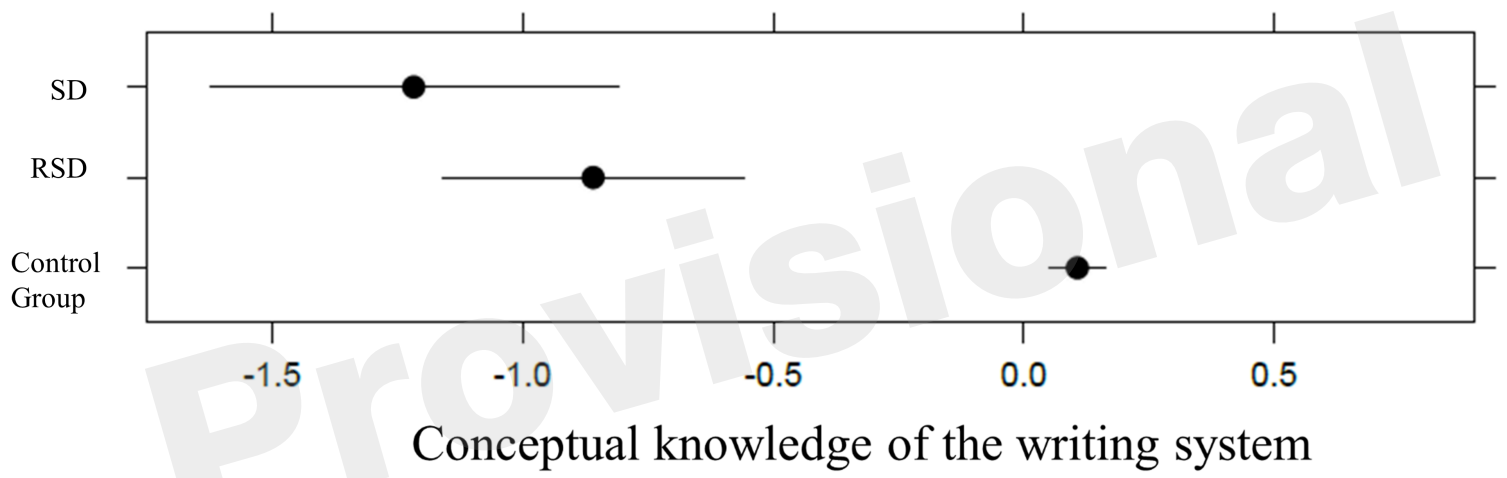


Figure 2.TIF

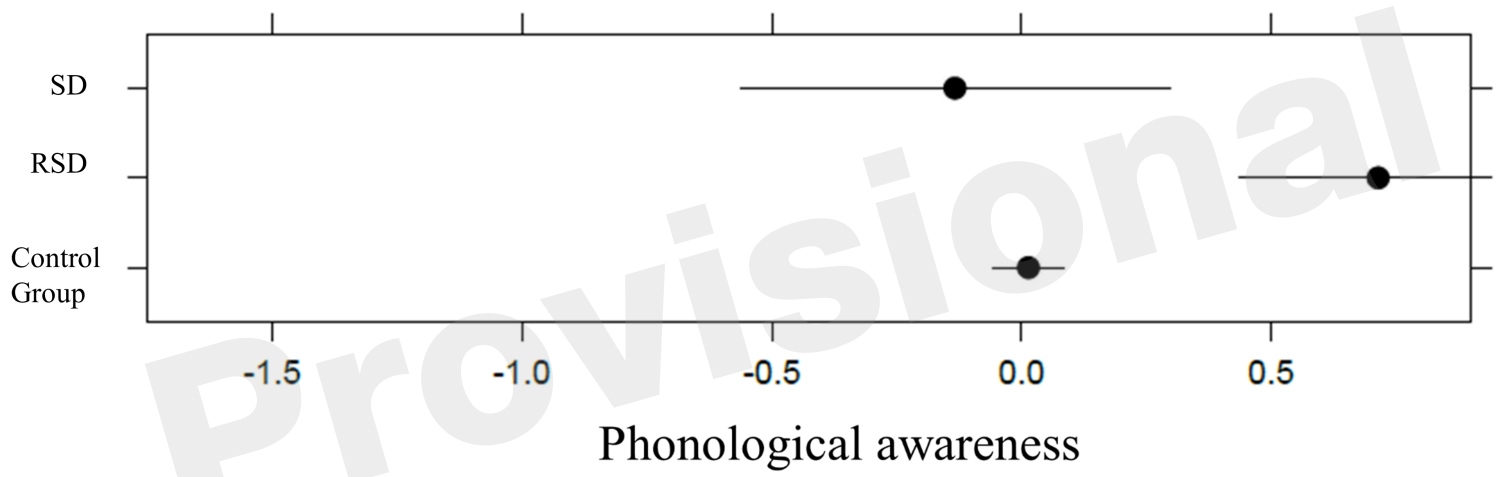


Figure 3.TIF

