

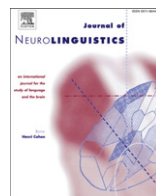


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Phonological or procedural dyslexia: Specific deficit of complex grapheme-to-phoneme conversion

Joël Macoir^{a,b,*}, Marion Fossard^c, Marie-Catherine Saint-Pierre^{a,d},
Noémie Auclair-Ouellet^{a,b}

^a Université Laval, Programme de maîtrise en orthophonie, Faculté de médecine, Département de réadaptation, 1050 avenue de la Médecine, Québec (Québec) G1V 0A6, Canada

^b Centre de recherche Université Laval Robert-Giffard, 2601 rue de la Canardière, Québec G1J 2G3, Canada

^c Université de Neuchâtel, Institut des sciences du langage et de la communication, Faculté des lettres et sciences humaines, Espace Louis Agassiz-1, CH-2000 Neuchâtel, Switzerland

^d Centre Interdisciplinaire de Recherche en Réadaptation et Intégration Sociale, 525 boulevard Hamel, Québec (Québec) G1M 2S8, Canada

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ABSTRACT

Phonological dyslexia is a written language disorder characterized by poor reading of nonwords when compared with relatively preserved ability in reading real words. There are two main theoretical proposals to explain this deficit: disruption of phonological processing or disruption to the nonlexical reading route affecting the grapheme-to-phoneme conversion rules (GPC). In this study, we report a single-case study of a mild aphasic patient with acquired phonological dyslexia. His ability was unimpaired for reading words, as well as in a wide range of tasks requiring the activation and explicit manipulation of phonological representations. He could also read every nonword with consistent GPC rules, whilst he was impaired for those with context-sensitive conversion rules, a pattern of performance never reported before. The implications of these results for theoretical explanations of phonological dyslexia are discussed, as well as the contribution of the patient's concomitant executive deficits to his performance in reading.

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* Corresponding author. Programme de maîtrise en orthophonie, Faculté de médecine, Département de réadaptation, Université Laval, Pavillon F-Vandry, Québec G1K 7P4, Canada. Tel.: +1 418 656 2131x12190; fax: +1 418 656 5476.

E-mail address: joel.macoir@rea.ulaval.ca (J. Macoir).

1. Introduction

Phonological dyslexia is a written language disorder characterized by poor reading of nonwords when compared with relatively preserved ability in reading real words. Since the first description of the syndrome by Derouesné and Beauvois (1979), there have been numerous reports of cases presenting with phonological dyslexia. Some authors (Friedman, 1996; Glosser & Friedman, 1990; Laine, Niemi, & Marttila, 1990) believe that phonological dyslexia is a mild variant of deep dyslexia, which is at the endpoint of the continuum of severity and is characterized by difficulties in reading nonwords and by the production of semantic errors in reading words. Between these two extremes are patients with a deficit limited to a slight impairment of nonword reading, patients with preserved word reading and a severe deficit for nonwords, and patients with an inability to read nonwords along with difficulty reading words (Berndt, Haendiges, Mitchum, & Wayland, 1996; Crisp & Lambon Ralph, 2006). Not only is there disagreement about the clinical characteristics of phonological dyslexia, there is also a lack of consensus in the literature about its functional origin.

The early reports of the impairment resorted to the 'dual-route' model of reading (Coltheart, 1978) to account for the deficit in reading nonwords. According to this model, which was implemented by Coltheart, Rastle, Perry, Langdon, and Ziegler (2001) in a computational model called the 'dual-route cascaded' model (DRC model), pronunciation of written words can be generated via two routes that function in parallel: a) a lexical route, in which the word's graphemes first map their corresponding representation in the orthographic input lexicon, which in turn can be used to directly activate the pronunciation of the whole word in the output phonological lexicon; and b) a nonlexical route, through the application of specific linguistic rules that convert each of the graphemes into their corresponding phonological representations (i.e. grapheme-to-phoneme rule-governed correspondences). Nonwords have no lexical correspondences and their pronunciation can only be generated via the nonlexical route. According to the model, the selective impairment in the application of GPC rules leads to phonological dyslexia.

Other researchers resorted to the connectionist 'triangle' model of reading (Harm & Seidenberg, 2001, 2004; Plaut, McClelland, Seidenberg, & Patterson, 1996) to account for acquired phonological dyslexia. In this model, reading aloud of both words and nonwords is achieved through the activation of recurrent connections between sets of orthographic, semantic, and phonological units. Nonwords are read by resorting to the connections established for words in the direct orthography-to-phonology pathway. As compared to familiar words, nonwords cannot activate the meaning units and cannot rely on established connections between orthography and semantics. As a consequence, they are more vulnerable to phonological impairment. By resorting to this model, Friedman (1995) identified two possible origins for acquired phonological dyslexia: (a) *impairment to phonological units* and/or (b) *impairment of direct connections between orthography and phonology*. According to Friedman, cases with *impairment to phonological units* (a) may stem from difficulties generating abstract phonological code or difficulties maintaining phonological units in auditory-verbal short-term memory. Therefore, in addition to difficulties in nonword reading, they should present with poor performance in nonword repetition as well as in tasks exploring the manipulation (e.g., phoneme and syllable blending, phoneme and syllable segmentation, syllable inversion) of phonology. A few patients (e.g., Berndt et al., 1996; Coslett, 1991) presenting with such a profile have been reported in the literature (For a review, see J.J. Tree, 2008). In the developmental domain, other researchers (e.g., Ramus Szenkovits, 2008; Szenkovits & Ramus, 2005) also suggested that phonological representations of dyslexic children are intact but that they presented with a deficit in accessing them.

On the other hand, according to Friedman (1995), because of the *impairment in the orthography-to-phonology direct pathway* (b), reading becomes highly dependent on the semantic route (i.e. orthography-to-meaning-to-phonology). Therefore, performance in reading directly depends on the richness or strength of semantic units (Friedman, 1996). Having no correspondence in the meaning units, nonwords should be particularly difficult to read. For similar reasons, closed-class words such as articles, conjunctions and pronouns, which carry little semantic information, should be read with less success than content words. For example, the performance in reading tasks of AN, the aphasic patient reported by Goodall and Phillips (1995), was affected for nonwords (30%) and closed-class words (48%), whereas content words were largely preserved (85%). More recently, Harm and Seidenberg (2001,

2004) considered that phonological dyslexia could only result from a generalized phonological impairment since the disruption of the orthography-to-phonology pathway they simulated in the computationally implemented version of the 'triangle' model led to difficulties not just in reading nonwords but also in reading irregular words.

In a systematic review of 38 published cases of phonological dyslexia, Tree (2008) recently reevaluated the validity of Friedman's criteria for distinguishing the two subtypes of the reading impairment. According to her survey, 7 of the 38 (18.5%) cases could be explained by a disruption of phonological processing (a), 5/38 cases (13%) could be explained by an impairment of direct connections between orthography and phonology (b), whilst the remaining cases (68.5%) fit neither of the two criteria (e.g., Caccappolo-van Vliet, Miozzo, & Stern, 2004; Manning & Warrington, 1995; Tree & Kay, 2006). It appears from all these studies that phonological dyslexia probably originated from different functional impairments. The presence or absence of a concomitant phonological deficit is certainly a key question in the debate concerning the underlying deficit leading to phonological dyslexia, as it is for the presence of reading difficulties of closed-class words associated with the deficit for nonwords.

The question of the contribution of the graphemic complexity of nonwords to the occurrence of phonological dyslexia has rarely been considered. In contrast with other languages like Italian or Spanish, French like English has a deep orthography, that is, there is no one-to-one mapping in phoneme-to-grapheme correspondences or grapheme-to-phoneme correspondences (GPC). For instance, the phoneme /o/ has different graphemic correspondences (EAU, AU, AUX, OS, OT, ...) and the grapheme 'EN' has many phonemic correspondences (e.g. /ã/ in enfant /ãfã/ 'child' and /ɛ̃/ in doyen /dwajɛ̃/ 'senior' or 'dean'). Different studies have shown that the graphemic complexity of words plays an important role in written word recognition. For example, Rey, Ziegler, and Jacobs (2000) observed that the time for detecting a letter in a word was longer when the letter was embedded in a multi-letter grapheme (e.g., A in BEACH) than when it corresponded to a single-letter grapheme (e.g. A in PLACE). Derouesné and Beauvois (1979) also showed such an effect on nonword reading in two of the four patients with acquired phonological dyslexia they reported. The performance of subjects A and B in their study was more affected for nonwords with a written form composed of three letters to read which required processing two of them together to derive a single phoneme (e.g., CAU → /k/ + /o/). Such a graphemic complexity effect in nonword reading was also reported by Howard and Best (1996) in a developmental phonological dyslexic subject. In a recent study about developmental dyslexia, Barca, Burani, Di Filippo, and Zoccolotti (2006) also showed a grapheme contextuality effect (i.e. better performance in reading words with simple graphemes than words with context-sensitive graphemes: in Italian, the pronunciation of sequences involving c and g is determined by context-sensitive rules and depends on the letters that follow). This effect was also reported in Italian in normal adults (Burani, Barca, & Ellis, 2006) as well as in normal children (Barca, Ellis, & Burani, 2007). However, to our knowledge, the effect of the nature and/or complexity of grapheme-to-phoneme correspondence (GPC) rules has never been considered in phonological acquired dyslexia.

In the present study, we report the case of FG, a French-speaking individual with acquired phonological dyslexia. The specificity of his reading problems, which affect only nonwords, allowed us to examine the various proposals on the functional origin of phonological dyslexia, as well as to demonstrate the importance of taking into account the nature and complexity of the GPC rules to explain the impaired processes in some cases of phonological dyslexia.

2. Method

2.1. Case description

FG is a 74-year-old right-handed man. He has a grade eleven education and worked as an auxiliary nurse. He had suffered from a chronic bipolar disease since 1982, with multiple episodes requiring many hospitalizations. He came to our attention in July 2005 for acute exacerbation of a bipolar disorder with suspected psychotic features requiring inpatient treatment. At admission, symptoms were compatible with manic exacerbation. Psychotic features were not confirmed. The Mini-Mental State (Folstein, Folstein, & McHugh, 1975) was administered to the patient, who obtained 24/30, a score within the

normal range (24–28). However, the examination revealed signs of his primary psychiatric disorder (exalted mood and paranoid suspicion). Moreover, an English-sounding foreign accent as well as mild agrammatism were noted. FG's past medical records reported the presence of this foreign accent in January 2003. It was first noticed at the psychiatric outpatient clinic consultation shortly after he was discharged from the inpatient service, which was required for manic exacerbation of his bipolar disorder in the fall of 2002. The presence of mild agrammatism was also recorded at that time.

Neuroimaging studies were performed while the patient was in euthymic condition (the reader will find MRI and PET pictures in Poulin, Macoir, Paquet, Fossard, & Gagnon, 2007). A magnetic resonance imaging (MRI) study including sagittal FLAIR and T2-weighted sequences and axial FLAIR, proton density, T1- and T2-weighted sequences was done in December 2005 using the standard protocol. The first interpretation was normal except for slight diffuse cerebral atrophy considered normal for his age. An 18F-fluorodeoxyglucose brain positron emission tomography was obtained with a dual-head coincidence camera (Vertex MCD-AC, Phillips). The reconstructed images showed diffuse hypometabolism in the frontal, parietal and temporal lobes bilaterally while the cerebellum, occipital lobe and subcortical structures were spared. There was also a focal deficit in the left anterior temporal lobe with prominence of the sylvian sulcus. When compared to the MRI, these deficits were related to asymmetric atrophy, which was retrospectively seen in the left temporal and frontal opercular/insular region (Poulin et al., 2007).

2.1.1. Neuropsychological evaluation

FG's performance on the neuropsychological tests, administered in March and April 2007, is shown in Table 1. Z-scores were based on the means and standard deviations of the control group. For each measure, any z-score ≤ 1.5 was considered impaired. Neuropsychological testing showed no impairment in tasks exploring orientation to time and space. FG's performance was normal on the task exploring concentration and selective attention (Symbol Digit Modalities Test; Smith, 1982). He showed good face recognition and presented no clinical signs of visual agnosia (BORB; Riddoch & Humphreys, 1993). There were no signs of unilateral neglect (Bells Test; Gauthier, DeHaut, & Joannette, 1989).

Praxis abilities were well preserved (PENO; Joannette et al., 1995). FG performed normally on tasks exploring episodic memory. His performance was within the normal range for the immediate story retelling subtest of the PENO battery (Joannette et al., 1995), for the two recalls of the DMS-48, a visual forced-choice recognition test (Barbeau et al., 2004), as well as for the pictorial recognition memory test and the short recognition memory test for faces (Camden Memory Tests; Warrington, 1996). The patient's short term memory was normal in the visuospatial modality on the Corsi block tapping test (Milner, 1971), whilst he presented with a mild deficit in the verbal modality (digit span). FG presented with deficits on every test exploring working memory and executive functions. He presented with a severe impairment on the interference condition of the Brown-Peterson task (Brown, 1958), a test that taps the ability to encode, maintain, and manipulate information in working memory (see Table 1). His performance on the color Stroop Test (Golden, 1978) showed abnormal sensitivity to interference. He obtained normal scores in the word reading and color naming conditions but his performance was impaired in the inhibition condition (i.e. color–word condition). FG had an abnormal performance on the Trail Making Test (Reitan & Wolfson, 1993). While in part A he was slow but had no errors, his performance was much poorer (very slow performance and numerous errors) on Part B in which he was asked to alternate between connecting numbers and letters in progressive sequential order (measure of mental flexibility). FG's performance was impaired (2 SD below the normal range) on the D-Kefs Tower Test (Delis, Kaplan, & Kramer, 2001), a complex task that measures the executive functions of spatial planning, rule learning, and inhibition of impulsive responding. Finally, FG's performance corresponded to low average on the Brixton spatial anticipation test (Burgess & Shallice, 1997), an instrument that measures the ability to detect rules in sequences of stimuli. In this task, most of the patient's errors consisted in the application of inadequate rules.

2.1.2. Language evaluation

With regard to language, speech output was fluent and well articulated, with no signs of word-finding difficulties. FG showed many characteristics usually reported for foreign accent syndrome (FAS): there were no signs of dysarthria (no slow, slurred, groping or labored articulation) or apraxia of

Table 1

Performance of FG and norms (mean and S.D. or range) on neuropsychological tests.

Test	FG's score	z-Score	Norm
Attention, working memory and executive functions			
Symbol digit modalities test	28	−0.54	33.31 (9.8)
Corsi block tapping test forward	5	−0.25	5.2 (0.8)
Corsi block tapping test backward	4	−0.82	4.9 (1.1)
Forward digit span	4	−1.36	5.5 (1.1)
Backward digit span	3	−0.83	4 (1.2)
Brown–Peterson test			
– Without interference	100%	0.37	98.33% (4.47)
– Mean of interference scores	42% ^a	−12.38	97.22% (4.46)
Stroop test			
– Color name reading	74 sec.	WNR	48.5 sec (25–86)
– Color naming	105 sec.	WNR	69.4 sec (46–123)
– Interference	249 sec. ^a	ONR	142.4 sec. (88–204)
Trail making test			
– Part A	61 sec.	−1.31	41.3 sec. (15)
– Part B	253 sec. ^a	−1.96	111.4 sec. (72.2)
Visual-perceptual tests			
Bells test (35)	32	−1.00	33.3 (1.3)
BORB			
– Length match task (30)	28	0.69	26.9 (1.6)
– Size match task (30)	27	−0.13	27.3 (2.4)
– Orientation match task (30)	24	−0.31	24.8 (2.6)
– Minimal feature view match (25)	25	0.85	23.3 (2.0)
– Foreshortened view task (25)	25	1.31	21.6 (2.6)
– Object decision – easy subtest (32)	30	−0.36	30.5 (1.4)
– Object decision – hard subtest (32)	25	−0.91	27.0 (2.2)
Motor control tests			
Pantomime imitation subtest (PEN0) (35)	29	−1.03	31.69 (2.6)
Arbitrary gesture imitation subtest (PEN0) (35)	33	0.29	32.54 (1.6)
Episodic memory			
Immediate story retelling subtest (PEN0) (23)	9	−1.01	11.62 (2.6)
DMS-48			
– First recall (48)	44	−0.87	46.08 (2.4)
– Second recall (48)	45	−0.81	46.56 (1.92)
Pictorial recognition memory test (30)	28	−0.39	28.6 (1.54)
Short recognition memory test for faces (25)	24	0.90	22.1 (2.1)

WNR = within the normal range; ONR = outside the normal range.

^a Indicates a z-score below the norm or outside the normal range.

speech (no dysfluency and no problems with phoneme sequencing) but acoustic analysis performed on speech samples recorded on digital audiotape showed the presence of abnormalities at the segmental and suprasegmental levels. Unfortunately, we had no premorbid recording of the patient's speech (Poulin et al., 2007). However, FG himself as well as one of his close friends, who has known him for over 30 years, confirmed that he never had this particular strange accent before its sudden appearance in January 2003. The patient also presented with mild expressive agrammatism. There were no phonemic or verbal paraphasias but speech was sometimes telegraphic with omissions of function words and grammatical bound morphemes as well as impoverished syntactic structure. FG's agrammatic speech was also characterized by a strong tendency to substitute clitic pronouns (which precede the verb in French) by their disjoint counterparts, leading to incorrect pronominalized structures (for a description of FG's agrammatism, see Macoir, Fossard, Nespoulous, Demonet, & Bachoud-Levi, 2010). Auditory input components assessed with the BECLA (Macoir, Gauthier, & Jean, 2005) were largely preserved (same vs. different judgment tasks on spoken syllables; lexical decision on spoken words).

Comprehension abilities at the lexical-semantic level (Pyramids and Palm Trees Test; Howard & Patterson, 1992) as well as at the syntactic-semantic level (Token test and MT-86; De Renzi & Faglioni, 1978; Nespolous et al., 1992) were normal (see Table 2).

Repetition was flawless for both words and nonwords (BECLA). Reading was canonical of phonological dyslexia with a preserved performance for words but impaired performance for nonwords. Written spelling of words and nonwords was impaired (BECLA). In written spelling to dictation, FG produced lexicalization errors (i.e. production of a word phonologically similar to the nonword) for nonwords while he exclusively produced phonological plausible errors (i.e. production of a response explicable in terms of the use of phoneme-grapheme conversion rules) for words, with a performance affected by orthographic regularity and lexical frequency. FG's performance was normal in confrontation naming (DO-80; Deloche & Hannequin, 1997) but he showed difficulties in letter and semantic category fluency tasks (Joanette et al., 1995) (see Table 2), a performance that could be attributed to the deficit in executive functioning. Finally, in a recent paper (Macoir et al., 2010), we also showed that FG presented with a procedural deficit affecting the application of rules in two linguistic domains (verbal and adjectival morphology, syntax) as well as in number processing whilst his ability to retrieve linguistic and numerical lexical representations was largely preserved. In summary, FG presented with a sudden onset of agrammatism, FAS, phonological dyslexia, and agraphia.

2.2. Control group

In all of the tasks in the experimental study, FG's performance was compared to the results of five (four in grapheme-to-phoneme conversion tasks) right-handed male controls matched for age (mean age = 74.8 years, SD = 0.84; modified *t*-test (Crawford & Howell, 1998) = -0.87 ; $p = 0.43$) and education level (mean education = 10.6 years, SD = 0.89; modified *t*-test = 0.41 ; $p = 0.7$). FG and the control subjects gave informed consent to participate in the study, according to the Declaration of Helsinki (BMJ 1991; 302:1194).

3. Experimental study

We investigated the nature of FG's phonological dyslexia through the administration of a wide variety of tasks. The experimental investigations are described in three sections: the first consisted of the background testing of reading abilities, the second section questioned the hypothesis of a phonological deficit, and the last section dealt with the hypothesis of a deficit in the application of grapheme-to-phoneme rules.

Table 2
Performance of FG and norms (mean and S.D. or range) on language tests.

Test	FG's score	z-Score	Norm
Language			
BECLA			
- Same-different judgment on spoken nonword pairs ($n = 36$)	30	-1.45	31.67 (1.15)
- Auditory lexical-decision ($n = 20$)	18	-0.58	19 (1.73)
- Repetition of words ($n = 25$)	25	1.72	24 (0.58)
- Repetition of nonwords ($n = 25$)	24	-0.27	24.17 (0.64)
- Reading of words ($n = 25$)	24	-0.29	24.17 (0.58)
- Reading of nonwords ($n = 25$)	15 ^a	-9.15	22.5 (0.82)
- Written spelling of words ($n = 36$)	24 ^a	-2.35	31.19 (3.06)
- Written spelling of nonwords ($n = 15$)	10	-1.70	13.17 (1.86)
Picture naming (DO-80) ($n = 80$)	72	-0.99	74.9 (2.94)
Letter fluency (PEN0)	5 ^a	-2.47	45.46 (16.4)
Category fluency (PEN0)	14 ^a	-3.45	47.85 (9.8)
Pyramids and palm trees test ($n = 52$)	47	-1.38	49.4 (1.74)
Token test ($n = 36$)	29	WNR	29–36
Spoken word/sentence-to-picture matching (MT-86) ($n = 47$)	44	-0.27	44.6 (2.19)
Written word/sentence-to-picture matching (MT-86) ($n = 12$)	12	1.47	10.81 (0.81)

WNR = within the normal range.

^a Indicates a z-score below the norm or outside the normal range.

3.1. Background testing of reading

3.1.1. Method

3.1.1.1. Material and procedure. FG and control subjects were asked to do various experimental tasks designed to explore reading abilities. Graphemic knowledge was assessed through the following four tasks: cross-case matching of letters (the subject was asked to select the allograph (e.g. R vs. B) corresponding to the presented letter (r); letter decision (the subject had to decide if the presented stimulus corresponded (e.g. f) or not (e.g. \$) to a letter of the alphabet); letter naming (the subject was asked to orally name the 26 letters of the alphabet); and mirror reading (stimuli were presented in a mirror-reversed manner and the subject was asked to read them aloud as fast as possible).

Knowledge about the association between graphemes and phonemes was assessed through the administration of the following two tasks: grapheme sounding (the subject was asked to select among two (e.g. m vs. d) the grapheme corresponding to the first letter of a nonword presented auditorily (e.g. dabète); and grapheme identification (the subject was asked to select among three (p vs. t vs. k) the grapheme corresponding to a phoneme (/t/) presented by the experimenter).

Access to the orthographic lexicon was assessed with a lexical decision task comprising 10 words and 10 nonwords.

With respect to word reading, FG and control subjects were asked to read aloud different experimental lists of words, presented in separate blocks in random order (stimuli were presented one by one on a computer screen using PowerPoint software) and controlled for concreteness (70 concrete and 70 abstract words), for grammatical class (37 closed-class words: pronouns, prepositions and conjunctions, and 37 open-class words: nouns, verbs and adjectives), for orthographic regularity (70 regular and 70 irregular nouns), for morphological complexity (prefixed nouns and adjectives, inflected verbs and adjectives and suffixed nouns and adjectives), and for length (1, 2, 3, and 4 syllable words). In all these lists, stimuli were also controlled for frequency.

The reading and comprehension of a text were assessed with two subtests (“Reading a text aloud” and “Reading comprehension of a text”) of the *Protocole Montréal-Toulouse d’examen linguistique de l’aphasie*–MT-86 (Nespoulous et al., 1992).

Finally, nonword reading was evaluated with an experimental list of 25 stimuli comprising letters with transparent GPC rules (i.e. each grapheme has only one unequivocal phonemic correspondence: A, O, I, L, M, R, ...), as well as some di- and tri-graphs with consistent GPC rules (e.g., the graphemes AU, EAU, OU, ON, ... are consistently pronounced /o/, /o/, /u/, /õ/, ...). These nonword stimuli were controlled for phonological complexity (12 simple CV syllables and 13 complex CCV and CVC syllables) and length (7 nonwords of 3–5 letters; 8 nonwords of 5–7 letters; 5 nonwords of 7–9 letters; 5 nonwords of 9–11 letters). Stimuli were presented one by one in random order on a computer screen using PowerPoint software.

3.1.1.2. Results. As shown in Table 3, the patient’s performance in reading tasks confirmed the presence of phonological dyslexia. He showed no difficulties in tasks assessing graphemic knowledge and knowledge of grapheme-phoneme associations or in the written lexical decision task. In word reading tasks, FG’s performance was largely preserved, showing no effect of concreteness, grammatical class, morphological complexity or length. Lexical frequency did not influence the patient’s performance. As compared to controls, FG’s performance was however slightly impaired (modified *t*-test: $t = -2.25$, $p < 0.05$) for irregular words (66/70), with the production of 3 regularization errors and 1 visual error.

The reading and comprehension of a text were flawless.

FG’s nonword reading was well below the mean of the control subjects (modified *t*-test: $t = -11.69$, $p < 0.001$), without influence of phonological complexity (simple syllables = 5/12; complex syllables = 7/13) or length (3–5 letters = 4/7; 5–7 letters = 4/8; 7–9 letters = 2/5; 9–11 letters = 2/5). All the errors resulted from the inappropriate application of grapheme-to-phoneme conversion rules (e.g., *nivon* → “nivonne” where the grapheme ON was treated as the sequence of two distinct graphemes O and N).

Table 3

Number and percentage of correct responses on word and nonword reading tasks for FG and mean (S.D.) for controls.

Task	FG	Controls
Graphemic knowledge		
– Cross-case matching of letters ($n = 26$)	26 (100%)	26 (-)
– Letter decision ($n = 60$)	60 (100%)	60 (-)
– Letter naming ($n = 26$)	26 (100%)	26 (-)
– Mirror reading ($n = 10$)	10 (100%)	9.8 (0.45)
Grapheme-phoneme associations		
– Grapheme sounding ($n = 20$)	20 (100%)	20 (-)
– Grapheme identification ($n = 20$)	20 (100%)	20 (-)
Written lexical decision ($n = 20$)	20 (100%)	19.67 (0.45)
Word reading		
– Concreteness		
■ Concrete ($n = 70$)	68 (97%)	69.4 (0.55)
■ Abstract ($n = 70$)	70 (100%)	70 (-)
– Grammatical class		
■ Open-class ($n = 37$)	37 (100%)	37 (-)
■ Closed-class ($n = 37$)	37 (100%)	37 (-)
– Orthographic regularity		
■ Regular ($n = 70$)	70 (100%)	70 (-)
■ Irregular ($n = 70$)	66 (94%)*	69.2 (1.3)
– Morphological structure		
■ Prefixed ($n = 15$)	14 (93%)	15 (-)
■ Suffixed ($n = 15$)	15 (100%)	15 (-)
■ Inflected adjectives ($n = 15$)	15 (100%)	15 (-)
■ Inflected verbs ($n = 15$)	15 (100%)	15 (-)
– Word length		
■ 1 syllable (3–5 letters) ($n = 30$)	29 (97%)	30 (-)
■ 2 syllables (5–7 letters) ($n = 30$)	30 (100%)	30 (-)
■ 3 syllables (7–9 letters) ($n = 30$)	30 (100%)	30 (-)
■ 4 syllables (9–11 letters) ($n = 30$)	30 (100%)	30 (-)
Nonword reading (25)	12 (48%)**	22.5 (0.82)

Difference between FG and controls (modified t -tests): *** $t = -11.69$, $p < 0.001$; * $t = -2.25$, $p < 0.05$.

3.2. Experiment 1 – hypothesis of a phonological deficit

3.2.1. Method

3.2.1.1. *Material and procedure.* FG presented with no deficits of phonology (no production of phonological errors in spontaneous speech, reading aloud, repetition). It is possible however that he had a deficit of this nature in more specific tasks and he was therefore tested with the following wide range of tasks requiring the activation and, for some tasks (i.e. phoneme and syllable blending, phoneme and syllable segmentation, syllable inversion), the explicit manipulation of phonological representations:

- a) Repetition: words (120 words controlled for frequency (60 words of high (mean = 150.59, range = 30–839) and low (mean = 10.32, range: 1–29) frequency (Baudot, 1992), length (30 words of 1, 2, 3 and 4 syllables), and syllable complexity (40 CV syllables and 80 complex (CVC, VC, CCV, ...) syllables), and nonwords (120 nonwords controlled for length (30 nonwords of 1, 2, 3 and 4 syllables), and syllable complexity (40 CV syllables and 80 complex (CVC, VC, CCV, ...) syllables) were pronounced by the examiner and the patient was asked to repeat them immediately.
- b) Rhyming tasks comprising: 1. Rhyme production (a bisyllabic nonword was auditorily presented (15 “easy” nonwords ending with a vowel and 15 “hard” nonwords ending with a consonant) and the patient had to produce a rhyming nonword); 2. Rhyme categorization (a bisyllabic target

- nonword (15 “easy” nonwords ending with a vowel and 15 “hard” nonwords ending with a consonant) was pronounced by the examiner, and the patient was asked to point to one of two written nonwords presented on a computer screen using PowerPoint software (the response and the distractor differed by the final vowel in “easy” stimuli; one of the 3 phonemes of the final syllable differed between the response and the distractor in “hard” stimuli) that rhymed with the spoken nonword); and 3. Rhyme judgment (two bisyllabic nonwords (15 “easy” nonwords ending with a vowel and 15 “hard” nonwords ending with a consonant) were auditorily presented and the patient had to indicate if they rhymed or not).
- c) Alliteration tasks comprising: 1. Alliteration production (a bisyllabic nonword was auditorily presented (15 “easy” nonwords beginning with a CV syllable and 15 “hard” nonwords beginning with a CVC syllable) and the patient had to produce a nonword beginning with the same syllable); 2. Alliteration categorization (a bisyllabic target nonword (15 “easy” nonwords beginning with a CV syllable and 15 “hard” nonwords beginning with a CVC syllable) was pronounced by the examiner, and the patient was asked to point on a computer screen to one of two written nonwords (both syllables differed between the response and the distractor in “easy” stimuli; one of the 3 phonemes of the first syllable differed between the response and the distractor in “hard” stimuli) that began with the same phoneme as the spoken nonword); and 3. Alliteration judgment (two bisyllabic nonwords (15 “easy” nonword pairs in which all the phonemes of the 1st syllable were identical or not and 15 “hard” nonword pairs in which the difference or similarity was on the 1st phoneme only) were auditorily presented and the patient had to indicate if they began with the same phoneme or not).
- d) Metaphonological tasks with phonemes comprising: 1. Phoneme blending (the examiner pronounced separately two phonemes (e.g., d-a) and the patient was asked to point on a computer screen to one of two written syllables corresponding to their blended form (da vs. ga); and 2. Phoneme segmentation (the patient was asked to point on a computer screen to one of two monosyllabic written nonwords corresponding to the heard nonword minus the final (e.g., fik: ki vs. fi) or the initial phoneme (e.g. kaf: fa vs. af).
- e) Metaphonological tasks with syllables comprising: 1. Syllable blending (the examiner pronounced separately three syllables (e.g., pro-si-fel) and the patient was asked to pronounce the nonword corresponding to their blended form (prosifel); 2. Syllable elision (the patient was instructed to segment by syllables (e.g., gra-fou-zi) the three syllable nonword pronounced by the examiner (grafouzi); 3. Syllable inversion (the patient was asked to repeat the bisyllabic nonword (e.g., friné) by reversing its syllables (néfri); and 4. Syllable length judgment (a stimulus was pronounced by the examiner and the patient was asked to indicate the number of its syllables) of spoken words and nonwords controlled for lexical frequency (words) as well as syllable complexity and length.

3.2.2. Results

As shown in Table 4, FG’s performance was at the same level as the control subjects for all of the tasks in the phonological processing battery. This result suggests that the functional origin of the patient’s reading deficit did not result from a phonological impairment (i.e. activation and explicit manipulation of phonological representations).

3.3. Experiment 2 – hypothesis of a deficit in the application of grapheme-to-phoneme conversion rules

3.3.1. Method

3.3.1.1. *Material and procedure.* In this experiment, FG and the controls were asked to read aloud two paired lists of 300 words and nonwords, controlled for letter (4–14 letters; mean = 6.83) and syllable (1–4 syllables; mean = 2.09) length, as well as for the nature and complexity of the following GPC contexts: 60 stimuli comprising letters with transparent GPC rules, 60 stimuli comprising di- and tri-graphs with

Table 4

Number and percentage of correct responses on phonological processing tasks for FG and mean (S.D.) for controls.

Task	FG	Controls
Repetition		
– Words (<i>n</i> = 120)	118 (98%)	116.4 (3.05)
– Nonwords (<i>n</i> = 120)	114 (95%)	114.6 (2.41)
Rhyming		
– Rhyme production (<i>n</i> = 30)	29 (97%)	29.8 (0.45)
– Rhyme categorization (<i>n</i> = 30)	28 (93%)	29.4 (1.34)
– Rhyme judgment (<i>n</i> = 48)	44 (92%)	45.2 (3.56)
Alliteration		
– Alliteration production (<i>n</i> = 30)	30 (100%)	29.8 (0.45)
– Alliteration categorization (<i>n</i> = 30)	28 (93%)	28.6 (3.13)
– Alliteration judgment (<i>n</i> = 40)	38 (95%)	38.8 (1.09)
Metaphonological tasks with phonemes		
– Phoneme blending (<i>n</i> = 30)	29 (97%)	29.6 (0.55)
– Phoneme segmentation		
■ Final phoneme (<i>n</i> = 20)	19 (95%)	19.8 (0.45)
■ Initial phoneme (<i>n</i> = 20)	19 (95%)	20 (–)
Metaphonological tasks with syllables		
– Syllable blending (<i>n</i> = 30)	30 (100%)	30 (–)
– Syllable segmentation (<i>n</i> = 30)	30 (100%)	30 (–)
– Syllable inversion (<i>n</i> = 30)	29 (97%)	29.8 (0.45)
– Syllable length judgment of spoken words		
■ 1 syllable words (<i>n</i> = 20)	20 (100%)	17.8 (2.17)
■ 2 syllable words (<i>n</i> = 20)	20 (100%)	18.8 (0.84)
■ 3 syllable words (<i>n</i> = 20)	20 (100%)	17.6 (2.3)
■ 4 syllable words (<i>n</i> = 20)	20 (100%)	19.75 (0.5)
– Syllable length judgment of spoken nonwords		
■ 1 syllable nonwords (<i>n</i> = 20)	19 (95%)	18.2 (2.49)
■ 2 syllable nonwords (<i>n</i> = 20)	19 (95%)	18.6 (3.13)
■ 3 syllable nonwords (<i>n</i> = 20)	19 (95%)	18.6 (1.52)
■ 4 syllable nonwords (<i>n</i> = 20)	18 (90%)	18 (3.39)

consistent GPC rules and, 180 stimuli with context-sensitive GPC rules (30 stimuli for the ‘vowel + S + vowel rule’ according to which the letter S, which is consistently pronounced /s/, changes to /z/ in an intervocalic position; 30 stimuli for the ‘C/G + E/I rule’ according to which the letters C and G, which are consistently pronounced /k/ and /g/, change to /s/ and /Z/ before the letters E and I; 120 stimuli for the ‘A, E, EU, OU + IL(L) rule’ according to which the graphemes I + L(L), pronounced /i + l = il/ according to transparent GPC rules, are consistently pronounced /j/ when preceded by A, E, EU and OU in the middle or at the end of words, a complex GPC process that results in the following correspondences: A + IL(L) = /aj/; E + IL(L) = /ɛj/; EU + IL(L) = /œj/; OU + IL(L) = /uj/). These contextual rules are productive in French. For example, according to New, Pallier, Brysbaert, and Ferrand (2004), there are 2410 words composed with the ‘A + IL(L) rule, and 6448 words composed with the ‘C/G + E/I rule’.

3.3.2. Results

As shown in Table 5, FG’s performance was normal for words, regardless of the nature and complexity of the GPC rules. As a whole, he showed difficulties with nonwords (words vs. nonwords: $\chi^2 = 57.25$, $p < 0.001$). However, he showed no difficulty with simple GPC nonwords, whilst his performance was substantially affected for nonwords with context-sensitive GPC rules.

The specific GPC was involved in 63 (91%) out of the 69 errors produced by FG on nonwords with the ‘A, E, EU, OU + IL(L) rule’. The errors consisted in: a) the inappropriate attachment of the grapheme ‘l’ to the preceding vocalic grapheme plus the application of the regular GPC to the grapheme ‘l’ (e.g., VANAIL → /vanɛl/ instead of /vanaj/), or the deletion of the ‘l’ (e.g., DRIFEILLONS /dri-fɛ-ɔ̃/ instead of /dri-fɛjɔ̃/); b) the omission of the grapheme ‘l’ and the application of the regular GPC to the grapheme ‘l’ (e.g., LAMAIL → /lamal/ instead of /lamaj/); and, in a few cases, c) the substitution of the complex

Table 5

Number and percentage of correct responses on reading tasks of words and nonwords according to the nature and complexity of GPC rules for FG and mean (S.D.) for controls.

Grapheme-to-phoneme rule	FG		Controls	
	Words	Nonwords	Words	Nonwords
Simple G-P correspondences ($n = 60$)	60 (100%)	58 (96%)	60 (-)	57.8 (0.84)
Simple di- and tri-graphs (AN, ON, EAU, ...) ($n = 60$)	59 (98%)	57 (95%)	57.6 (0.89)	57.4 (1.14)
Contextual G-P correspondences				
– Vowel + S + Vowel ($n = 30$)	30 (100%)	8 (27%)*	30 (-)	29.25 (1.5)
– C + I, E ($n = 15$)	15 (100%)	12 (80%)	15 (-)	14.8 (0.45)
– G + I, E ($n = 15$)	14 (93%)	12 (80%)	15 (-)	14.6 (0.56)
– Ending with AIL ($n = 15$)	15 (100%)	5 (33%)*	15 (-)	15 (-)
– with AILL in the middle ($n = 15$)	13 (87%)	6 (40%)*	15 (-)	15 (-)
– Ending with EIL ($n = 15$)	15 (100%)	1 (7%)*	15 (-)	14.75 (0.5)
– with EILL in the middle ($n = 15$)	12 (80%)	9 (60%)	15 (-)	15 (-)
– Ending with EUIL ($n = 15$)	15 (100%)	8 (53%)*	15 (-)	15 (-)
– with EUILL in the middle ($n = 15$)	14 (93%)	6 (40%)*	15 (-)	15 (-)
– Ending with OUIL ($n = 15$)	15 (100%)	8 (53%)*	15 (-)	14.5 (1)
– with OUIILL in the middle ($n = 15$)	15 (100%)	8 (53%)*	15 (-)	15 (-)
– Total contextual G-P corresp. ($n = 180$)	173 (96%)	83 (46%)*	180 (-)	177.9 (3.01)
Total performance ($n = 300$)	292 (97%)	228 (69%)*	297.6 (0.89)	293.1 (4.99)

Difference between words and nonwords (chi-square tests): * $p \leq 0.05$; ** $p \leq 0.01$; *** $p \leq 0.001$.

grapheme by another of the same type (e.g., FÊTEIL \rightarrow /fetɛj/ instead of /fetɛj/). The remaining errors involved transparent GPC rules. For the ‘vowel + S + vowel rule’ as well as for the ‘C/G + E/I rule’, all of the errors produced resulted from the non-application of the context rule and the application of the simple GPC (e.g., LASATE \rightarrow /lasat/ instead of /lazat/; FAGI \rightarrow /fagi/ instead of /fazi/).

4. Discussion

FG is a patient with aphasia characterized by agrammatism, FAS, and agraphia. He also presented with phonological dyslexia and we conducted an experimental study to investigate the nature and origin of this deficit. We first addressed the hypothesis of a phonological deficit through the administration of a wide range of tasks requiring the activation and the explicit manipulation of phonological representations. FG’s performance was at the same level as the control subjects for all of these tasks, a result that suggests that phonological impairment was not the functional origin of his reading deficit. The hypothesis of a deficit in the application of grapheme-to-phoneme conversion rules was then explored through reading tasks of words and nonwords. In these tasks, FG was affected for nonwords but his performance was directly linked to the nature and complexity of the GPC rules. He was at the control level for simple GPC nonwords, whilst he was substantially impaired for nonwords with context-sensitive GPC rules. As a whole, this pattern of performance directly challenges current theoretical models of reading.

4.1. FG and the ‘triangle’ model of reading

In the Introduction to this article, we briefly presented the two potential loci of impairment proposed by Friedman (1995) to account for phonological dyslexia. By resorting to the ‘triangle’ model of reading, she suggested that poor nonword reading can be explained either by (a) impairment to phonological units; and/or (b) impairment of direct connections between orthography and phonology. According to Friedman, cases with impairment to phonological units (a) presented with difficulties not just in nonword reading but also in nonword repetition, as well as in every task exploring the manipulation of phonology. Friedman (1995) also suggested the existence of a variant of this profile in which patients presented with poor auditory-verbal short-term memory leading to difficulty maintaining phonological codes. We have shown that FG was at the control level for the repetition of nonwords up to 4 syllables, as well as in every task requiring the explicit retention and manipulation of

phonological representations in short-term memory. FG's performance however was affected in tests exploring working memory and executive functions and we cannot completely rule out the possibility that this impairment could contribute to his phonological deficit.

With respect to the second profile (b), Friedman (1995) suggested that, because of impairment in the orthography-to-phonology direct pathway, reading becomes highly dependent on the semantic route (i.e. orthography-to-meaning-to-phonology). Patients with this particular profile of phonological dyslexia presented with substantial difficulties in reading nonwords since they have no correspondence in the meaning units. These patients should also present with difficulties in reading closed-class words such as articles, conjunctions and pronouns, since these words carry little semantic information. Harm and Seidenberg (2001, 2004) simulated the disruption of the orthography-to-phonology pathway in the computationally implemented version of the 'triangle' model. According to them, phonological dyslexia could only result from a generalized phonological impairment since this disruption led to difficulties not just in reading nonwords but also in reading irregular words. In the present paper, we have shown that FG was largely unimpaired for words in general, including irregular words and closed-class words.

In summary, FG's pattern of performance is not consistent with the predictions of the 'triangle' model of reading. He had no phonological impairment and was impaired for nonwords only. Moreover, his performance for nonwords was directly linked to the nature and complexity of the GPC rules, a pattern of performance which can be explained by resorting to the DRC model of reading (Coltheart et al., 2001).

4.2. FG and the DRC model of reading

According to the DRC model of reading (Coltheart, Curtis, Atkins, & Haller, 1993; Coltheart et al., 2001), the selective impairment of the nonlexical route, as observed in FG, leads to phonological dyslexia. This reading route involves three successive processing stages: 1) graphemic parsing, 2) GPC, and 3) phoneme blending. The graphemic parsing is responsible for converting letter strings into graphemes. In the GPC process, the graphemes are translated to the corresponding phonemes, one by one, from left to right (Coltheart, 1978). According to Caramazza and Miceli (1990), at least three variables may determine the selection of the GPC rules: the relative frequency of the grapheme-to-phoneme mapping, the within-syllable positional constraint, and the contextual constraints. In French, some GPC rules are dependent on the context. For these specific rules, the pronunciation does not derive from a simple grapheme-to-phoneme mapping but is directly dependent on the environment in which specific graphemes are presented. For example, when the graphemes I + L(L) are preceded by the graphemes A, E, EU and OU in the middle or at the end of words, they are consistently pronounced /j/, resulting in the following correspondences: A + IL(L) = /aj/ ; E + IL(L) = /ɛj/; EU + IL(L) = /œj/; OU + IL(L) = /uj/. In every other context, the same association of graphemes (I + L)L is pronounced /i + l = il/ according to transparent GPC rules. Finally, the stage of phoneme blending consists in the integration of each of the phonemes derived from the GPC rules into a unified phonological form.

A deficit in graphemic parsing usually manifests by the incorrect assignment of phonemes to letters instead of graphemes. In fact, patients with this type of deficit convert each letter into the corresponding phonemes before blending them into graphemic representations. This stage of the nonlexical route of reading is unimpaired in FG. He encountered no problems for simple, term-by-term, non-contextual, GPC rules or for di- and tri-graphs GPC rules. As discussed above, FG had no problem manipulating phonology and a deficit at the stage of phoneme blending is therefore highly unlikely. However, FG presented with a specific deficit functionally localized at the GPC stage. More specifically, his performance was affected only when he had to read nonwords requiring the application of contextual complex rules. The effect of the graphemic complexity of written stimuli on nonword reading was observed by Derouesné and Beauvois (1979) in two patients with acquired phonological dyslexia, by Howard and Best (1996) as well as by Barca et al. (2006) in developmental dyslexia, by Burani et al. (2006) and by Barca et al. (2007) in Italian in normal adults and normal children respectively. To the best of our knowledge, however, FG is the first reported case with acquired phonological dyslexia in which the impairment was directly linked to the nature and complexity of the GPC rules.

In another study conducted with FG (Macoir et al., 2010), we showed that he was impaired when he was required to apply inflection rules to nonverbs and nonadjectives, whilst he was unimpaired for real verbs and real adjectives. In the present study we have shown that the patient's deficit in reading was also limited to nonwords. Instead of a deficit in rule application, another explanation could be that the patient's reading deficit for nonwords originates from his executive disorders affecting controlled cognitive processes (i.e. novel problem solving, shifting of mental sets, inhibition of prepotent or previous responses). That would be less the case for non-contextual GPC rules which would be processed more automatically, thus demanding less cognitive resources. In fact, the patient presented no difficulty when he could apply term-by-term GPC rules or when the grapheme that immediately follows an ambiguous grapheme (e.g., 'C/G + E/I rule') allows disambiguating the graphemic context. For other contextual GPC rules, the pronunciation can only be derived by taking into account a wider portion of the graphemic environment ('vowel + S + vowel rule' and 'A, E, EU, OU + IL(L) rule'), which therefore could require more executive resources. Rey et al. (2000) also proposed a similar explanation to account for the additional time required to read nonwords with multi-letter graphemes as compared to nonwords with single-letter graphemes. According to them, for a nonword with a multi-letter grapheme such as VANAIL, activation will spread to the graphemes V, A, N, A, I, AI, and L. The multi-letter grapheme AI will compete with the single-letter graphemes A and I. This competition has a processing cost that is not required for reading nonwords composed only of single-letter graphemes. Based on our study with FG, we suggest that for nonwords with contextual GPC rules, as in the former example, the reader will also have to take into account the context in which the multi-letter appears (AI + L) to derive the correct pronunciation, resulting in an even more costly processing load.

5. Conclusion

In FG, the asymmetric atrophy found in the left temporal and frontal opercular/insular region was highly consistent with neuroanatomical findings about reading processes. According to various studies (e.g., Paulesu et al., 2001; Pugh et al., 2001) the left frontal operculum, the adjacent anterior insula, and the left lateral inferior parietal cortex/posterior superior temporal cortex are brain regions with higher activation for reading pseudowords compared to real words. More specifically, the study by Fiez, Balota, Raichle, and Petersen (1999) revealed that left fronto-insular activations were associated with the nonlexical route of reading. Using event-related fMRI to investigate visual word and pseudoword recognition, Fiebach, Friederici, Müller, and Cramon (2002) also showed that the grapheme-to-phoneme conversion was sustained by the left inferior frontal gyrus and the anterior insula. However, these brain areas are also largely involved in working memory and executive functions (Derrfuss, Brass, Neumann, & von Cramon, 2005; Graves, Desai, Humphries, Seidenberg, & Binder, 2010; Grosbras, Laird, & Paus, 2005) as well as in the executive control mechanisms for rule application (Koechlin & Jubault, 2006). Moreover, different studies have also reported the involvement of the insular region, impaired in FG, in rule learning and rule application (Seger & Cincotta, 2005; Ullman & Corkin, 1997; Ullman et al., 2005). From a neuroanatomical viewpoint, it is therefore difficult to disentangle neural systems devoted to nonlexical reading processes from those sustaining executive functions and rule application.

In closing, we have shown that FG presented with a profile of phonological dyslexia never reported before, exclusively affecting nonwords with contextual complex GPC rules. The extent to which the application of these rules relies on linguistic and/or executive processes remains unclear. Along with Nickels and her colleagues (Nickels, Biedermann, Coltheart, Saunders, & Tree, 2008), we argue that phonological dyslexia can be the result of various impairments. Some patients presented with clear phonological deficits whereas others, like FG, showed specific deficits exclusively affecting the nonlexical reading route. As a whole, our data underline the need for studies in which the nature and specificity of the GPC rules are controlled in order to clarify the issue of the independence of the nonlexical reading route compared to phonological processes. The question related to the application of these rules on the one hand, and the executive and controlled processes on the other, should also be addressed in further studies.

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