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Acquired surface dysgraphia and dyslexia in the semantic variant of primary progressive aphasia: a single-case study in Spanish

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ABSTRACT

Background: The diagnostic criteria for the semantic variant of primary progressive aphasia (svPPA) include the possible presence of acquired surface dyslexia or dysgraphia. Acquired surface dyslexia is characterized by a greater difficulty in reading irregular words with the production of regularization errors. On the other hand, acquired surface dysgraphia is characterized by difficulties in writing irregularly spelled words, which are stimuli that produce phonologically plausible errors. The identification of these patterns in Spanish has been subject to controversy due to the orthographic transparency of the language and its lack of irregular words. However, differences do exist between reading (completely transparent) and writing (considerably irregular) and thus, designing tests to identify acquired surface dysgraphia is a simple task, since there are irregular words for writing in Spanish. Nevertheless, few cases of acquired surface dysgraphia have been reported in Spanish-speaking patients. In addition, the identification of acquired surface dyslexia in Spanish-speaking patients requires the use of tasks other than reading irregular words.
Aims: The aim of this paper is to report the reading and writing impairments of a Spanish-speaking patient with svPPA, and show that it is possible to identify patterns of acquired surface dysgraphia and dyslexia in a transparent language.
Methods and procedure: Single case study of a Spanish-speaking patient with svPPA. The tests administered were: writing to dictation of words (both regular and irregular) and nonwords, reading of words and nonwords (measuring accuracy and reaction times) and lexical decision with pseudohomophones.
Outcomes and results: We found a regularity effect in the writing to dictation task with the generation of several phonologically plausible errors, a loss of lexical advantage in reading latencies although reading accuracy was conserved, and a pseudohomophone disadvantage

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effect in the lexical decision task; thus, resulting in a profile consistent with the patterns of acquired surface dysgraphia and dyslexia.
Conclusions: The results obtained indicate it is possible to identify these patterns when the characteristics of the language are taken into consideration in the selection of tests to be administered. Therefore, it is possible to test for all the established criteria for svPPA in Spanish-speaking patients.

1. Introduction

Primary progressive aphasia (PPA) is an insidious-onset, gradually progressive neurodegenerative disorder whose core features are the initial impairments of the language domain (Mesulam, 2001). Three main variants or forms of PPA are recognized according to the language impairment pattern of the patient: agrammatic, semantic and logopenic PPA (Gorno-Tempini et al., 2011). The consensus clinical criteria for the identification of semantic variant primary progressive aphasia (svPPA) includes two mandatory requirements: impaired confrontation naming (anomia) and impaired single-word comprehension. In addition, three of the following four conditions must be met: impaired object knowledge, particularly for low-frequency or low-familiarity items; acquired surface dyslexia or dysgraphia; spared repetition; and/or spared speech production. Imaging-supported criteria include the presence of predominant anterior temporal lobe atrophy in CAT or MRI scans and/or hypoperfusion or hypometabolism in the same region in PET or SPECT scans. In addition to meeting the clinical criteria, definite pathologic diagnosis requires histopathologic evidence of either a specific neurodegenerative pathology (for example: FTLD-tau, FTLD-TDP, Alzheimer disease, etc.) or the presence of a known pathogenic mutation (Gorno-Tempini et al., 2011).

Using the consensus diagnostic criteria for svPPA presents cross-cultural difficulties due to the particularities of different writing systems. This is because the diagnostic criteria include the presence of acquired surface dyslexia or dysgraphia (Gorno-Tempini et al., 2011), patterns that cannot be identified in the same manner in transparent writing systems, such as Spanish or Italian, as in opaque writing systems, such as English or French.

In the context of the dual route models of reading and writing words (Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001; Ellis & Young, 1988; Houghton & Zorzi, 2003), acquired surface dyslexia and dysgraphia are interpreted as pathological patterns that reflect greater difficulty using the lexical routes of reading and writing, with the relative preservation of the respective non-lexical routes and a performance that is overdependent on them. In short, the dual route models assume a functional architecture in which, following an initial analysis of the stimulus in the visual analysis system (if it is a written word) or auditory analysis system (in the case of a word that is heard), processing then takes one of two possible routes. The lexical route operates through associating the orthographic and phonological representations of words, and thus can only be used for the reading or writing of known words, no matter if they are regular or irregular. This route consists of three key components: the input lexicon (orthographic or phonological, depending on whether the stimulus is visual or auditory); the semantic system (where the meaning of words is stored); and the output lexicon (phonological or orthographic, again, depending on whether the task is reading aloud or spelling to dictation). Likewise, there is a second, non-lexical route – also known as the phonological route – which operates step by step using sublexical conversion systems: grapheme-to-phoneme in reading, and phoneme-to-grapheme in writing. This route allows the reading and spelling to dictation of both non-words and unknown words, as well as regular words. However, when this route processes irregular words, two types of characteristic errors arise: regularization errors (in reading) and phonologically plausible errors (in dictation).

Acquired surface dyslexia is a reading impairment that is characterized by greater difficulty in reading irregular words, stimuli with which patients produce regularization errors (English examples: STEAK > /stik/; FLOOD > /flud/, extracted from Patterson, Marshall, & Coltheart, 2017) while the reading of regular words and nonwords is relatively preserved (Marshall & Newcombe, 1973; Patterson et al., 2017). In the context of the dual route model, acquired surface dyslexia is interpreted as the result of an impairment of any of the components of the lexical route while the non-lexical route remains relatively preserved. There are at least three possible loci of impairment in the lexical route: one in the access to or in the orthographic input lexicon itself; a second in the access to the semantic system from the orthographic input lexicon, or in the semantic system itself; and a third in the access to or in the phonological output lexicon itself (Ellis, Lambon Ralph, Morris, & Hunter, 2000; Humphreys & Evett, 1985).

In acquired surface dysgraphia, patients demonstrate greater difficulties in writing irregularly spelled words (Beauvois & Dérouesné, 1981; Roeltgen & Heilman, 1984), stimuli that produce the phonologically plausible errors that are characteristic of the condition (Spanish examples: /siya/ [SILLA] (*chair*) > CILLA /siya/; /beso/ [BESO] (*kiss*) > VESO /beso/; English examples: BURY > BERY; COUGH > KOF), while the writing to dictation of nonwords or regular words is much more well-preserved. In the context of the dual route model, acquired surface dysgraphia is interpreted as the result of an impairment of the lexical route of writing.

In both acquired surface dyslexia and dysgraphia, patients preserve reading or writing abilities through the non-lexical route, that is, through the grapheme-to-phoneme conversion process for reading, and the phoneme-to-grapheme conversion process in dictation. This allows them to correctly execute the reading and writing to dictation of both nonwords and regular words, which is particularly important for interpreting the performance of Spanish-speaking patients, as all Spanish words are completely consistent words, i.e., they can be pronounced correctly by applying grapheme-to-phoneme rules. On the other hand, patients with acquired surface dyslexia and dysgraphia appear to have lost lexical orthographic information and, as a result, fail to read and write irregular words, i.e., arbitrarily spelled words of different degrees of inconsistency. The use of residual sublexical ability to read or write irregular words causes regularization errors in reading and phonologically plausible errors in writing.

The PPA consensus includes suggested tests to assess the performance and symptoms specified in the diagnostic criteria (Gorno-Tempini et al., 2011). Lists of regular and irregular words, in which variables such as length, frequency and word class are controlled, are recommended for the evaluation of reading and writing. This specification is easily satisfied in opaque writing systems, which have many irregularly spelled words for reading and writing. However, it is difficult to implement in languages with transparent orthography, as they lack irregular words altogether or have very few of them.

Spanish is one of the orthographies considered transparent. Nevertheless, there is a difference between reading and writing, as sublexical heterographic homophony is one of the characteristics of the Spanish orthographic system. Some Spanish phonemes can be written in two, or at most three, different ways. For example, the phoneme /b/ is written using the letters B or V in different words: /beso/ > BESO (*kiss*); /baca/ > VACA (*cow*); the phoneme /s/ is written S or C (before E or I) or Z[1]: SOMBRA > /sombra/ (*shadow*), CIELO > /sielo/ (*sky*), ZAPATO > /sapato/ (*shoe*); the phoneme /x/ is written J or G before E or I: JEFE > /xefe/ (*boss*), GESTO > /xesto/ (*gesture*); the letter H preceding any vowel is silent, it is not pronounced[2]: /umo/ > HUMO (*smoke*), /ielo/ > HIELO (*ice*), etc. Unlike opaque languages, Spanish lacks heterophonic homographs. It does not have sequences of letters that are pronounced differently depending on the word in which they are found. This means that any word can be read correctly by applying grapheme-to-phoneme rules. It does not matter whether a word is written with B or V; as long as one understands the rule, "B and V are pronounced /b/", the word can be pronounced correctly. Likewise, should a letter H appear, the rule indicates, "if an H comes before a vowel, it is silent.". As Funnell (2013) stated, it is not possible to build a list of irregular words for reading in Spanish. However, the situation is completely different for writing. When writing dictated words that contain the phonemes /b/, /s/, /x/, the sublexical rules of phoneme-to-grapheme conversion are not sufficient to be able to write the words correctly; the lexical orthography of the words must be known. For example, in order to write /beso/ > BESO (*kiss*) correctly, the lexical orthography of the word must be known, as the sublexical rule /b/ > B or V is insufficient and could lead to the production of a phonologically plausible error, such as VESO. Likewise, lexical orthographic knowledge is necessary to be able to correctly write the words /baca/, /sielo/, /xefe/, /xesto/, /ielo/ > VACA, CIELO, JEFE, GESTO, HIELO (*cow, sky, boss, gesture, ice*); the sublexical rules of phoneme-to-grapheme conversion could produce phonologically plausible errors, such as BACA, SIELO, GEFE, JESTO and IELO, respectively.

The difference that exists in Spanish between reading (completely transparent) and writing (considerably irregular) was not taken into account in the Frontotemporal Lobar Degeneration (FTLD) Module of the Uniform Data Set (UDS), one of the most important protocols for the cross-cultural study of PPA. In fact, the Spanish version of the Neuropsychological Battery of the FTLD Module (National Alzheimer's Coordinating Center & FTLD work group of the ADC Program, 2015. Spanish version from: Grupo de Trabajo Latino del Programa ADC y la Red Latinoamericana de Demencias, 2015) omits the writing and reading tests, which significantly reduces the possibility of diagnosing svPPA in Spanish; recall that, as previously mentioned, the criteria states that three of four conditions must be present, one of which is acquired surface dyslexia or dysgraphia.

Carrying out tests to identify acquired surface dysgraphia is a relatively simple task since, as we have just shown, there are irregular words for writing in Spanish. Nevertheless, little attention has been paid to the study of acquired surface dysgraphia in Spanish-speaking patients; few cases have been reported, and all of these have been in the context of a focal lesion (Ferreres, López, & Fabrizio, 2012; Ferreres, Martínez Cuitiño, & Olmedo, 2005; García Orza, Lazcano, & Alvarez, 2002; Iribarren, Jarema, & Lecours, 2001). Though they are few, these studies suggest that it is possible to use writing to dictation (of regular and irregular words) with Spanish-speaking patients to identify phonologically plausible errors and diagnose a pattern of acquired surface dysgraphia. This would allow the suggestions raised in the svPPA diagnostic consensus (Gorno-Tempini et al., 2011) with regard to the criterion for acquired surface dysgraphia to be followed in Spanish.

Identifying acquired surface dyslexia in Spanish speakers is more problematic, as all words in Spanish are transparent for reading and patients can read all words through grapheme-to-phoneme conversion, without resorting to lexical orthographic information (Ferreres & López, 2014; Valle-Arroyo, 1996). This makes it impossible to observe regularization errors in reading, which is the pathognomonic symptom of acquired surface dyslexia, since it is not possible to regularize that which is already regular (in reading). In another transparent language, Italian, the very small number of irregularities in reading are exploited to assess for regularization errors. For example, Ripamonti, Lucchelli, Lazzati, Martini, and Luzzatti (2016) used the task of reading words with unpredictable stresses aloud. In Italian, it can only be determined through lexical orthographic knowledge whether a trisyllabic word is stressed on the penultimate or antepenultimate syllable. The stress position is not marked diacritically, nor can it be inferred through rules. Nevertheless, this resource is not available in Spanish, as there is no ambiguity in terms of the position of the stress. Matías-Guiu et al. (2017) tried to overcome this difficulty in Spanish-speaking patients by using tests of reading foreign words and reading words with the accent omitted. However, the usefulness of reading foreign words has been questioned, as there is such a large variation in the pronunciation of foreign words by the population (Ferreres et al., 2005; Matías-Guiu et al., 2017). Meanwhile, the task of reading words with the accent omitted may not be sufficiently sensitive, since the position of an accent can be inferred from obtaining the phonemes of a word (Ferreres et al., 2005).

Though they may not be equivalent to the irregular word reading test, there are other tests that can show spared sublexical reading in patients who have lost their lexical reading abilities (Coltheart, 1982). These are the homophone comprehension, visual lexical decision with pseudohomophones (PSH) and reading latencies tests (Valle-Arroyo, 1996). In the homophone comprehension test, the patient must choose which of two homophonic words corresponds to the definition of, for example, “a ripple on the surface of water”, where the subject must choose between OLA /ola/ (*wave*) and its homophone HOLA /ola/ (*hello*). The task requires the subject to have preserved orthographic information, and it cannot be compensated by reading through grapheme-to-phoneme conversion. However, this task is not suitable for a population of patients with svPPA, as their semantic impairments affect their performance in any comprehension task, making it difficult to determine if task failure is due to difficulties in lexical orthographic information or semantic difficulties. The lexical decision with PSH test uses heterographic homophony to construct stimuli that “sound” like words but are written incorrectly. For instance, VOCA, NERBIO and VESO are the PSH for the words BOCA (*mouth*), NERVIO (*nerve*) and BESO (*kiss*). If a patient says that PSH are real words (false positive errors), it strongly suggests there is a loss of lexical orthographic information and that the lexical decision is being made through phonological mediation (Coltheart et al., 2001). The fact that PSH may be accepted as real words in the lexical decision task can be useful in identifying acquired surface dyslexia in Spanish speakers. Finally, the reading latency measurement test allows the observation of whether the patient has a lexical advantage (shorter latencies for words than nonwords). The lexical advantage can be observed in expert readers, due to the greater efficiency of lexical reading compared to reading through grapheme-to-phoneme conversion. The loss of lexical advantage in reading can be attributed to the patient reading words and nonwords through grapheme-to-phoneme conversion. The three tests mentioned above have been used to identify acquired surface dyslexia in Spanish-speaking patients with focal lesions (Ferreres et al., 2005, 2012; for a review of the study of acquired dyslexias in Spanish, see Ferreres & López, 2014) and, as stated, the latter two tests could be administered for identifying patients with svPPA. It is also possible to observe failures in these same tasks in English-speaking patients with acquired surface dyslexia (Patterson et al., 2017), however, due to the ease with which regularization errors can be detected, the tasks have been used more for identifying the specific locus of impairment within the lexical route of reading. In Spanish, measuring word and nonword reading times can provide information regarding the functioning of the lexical and non-lexical routes, but it does not make it possible to identify which of the components is compromised. Meanwhile, errors in the homophone comprehension and lexical decision with PSH tasks suggest the presence of input acquired surface dyslexia, that is, acquired dyslexia in which the impairment is located in the access to or in the orthographic input lexicon itself. Thus, the findings in Spanish that make use of the aforementioned instruments refer to the identification of the type of acquired surface dyslexia with a compromise of the lexical orthographic information (Ellis et al., 2000; Humphreys & Evett, 1985).

Since Gorno-Tempini et al. updated the diagnostic criteria in 2011, there have been very few studies of reading and writing in Spanish-speaking patients. Wilson and Martínez-Cuitiño (2012) studied a patient with semantic dementia but did not find acquired surface dyslexia and, although the study did not focus on writing, they mentioned in the general assessment of language production that the patient produced phonologically plausible errors in writing. Matías-Guiu et al. (2017) studied reading in five patients with svPPA, using tests of reading foreign words and reading words with the accent omitted. They encountered difficulties with the latter test and questioned the usefulness of the former. Performance in writing tasks was not reported in their paper.

Taking into consideration the context and particularities of the Spanish writing system, it is important to determine which tests can be administered for a reliable identification of acquired surface dysgraphia and dyslexia patterns and, thus, allow the full application of the international consensus criteria for the diagnosis of svPPA in Spanish-speaking patients.

The aim of this paper is to present the study of a Spanish-speaking patient with svPPA who presented evidence of both acquired surface dysgraphia, through tests of spelling to dictation of regular and irregular words and nonwords, and acquired surface dyslexia, through tests of lexical decision with PSH and the measurement of reaction times (RT) in reading.

2. Materials and method

This study has been approved by the Ethics Committee of the Faculty of Psychology, University of Buenos Aires. Written informed consent was obtained from all participants of this investigation in accordance with the Helsinki Declaration.

2.1 Participants

2.1.1 Patient BF

BF is a 71-year-old woman who is a right-handed, native Rioplatense Spanish speaker with 13 years of education, retired teacher and resident of the province of Buenos Aires, Argentina. In her working life, BF was a grade school teacher and was always interested in the arts and reading. She was a skilled pianist and drawer, activities she continues to practice though she encounters certain difficulties now; though she can produce a highly-detailed copy, she cannot make spontaneous drawings nor draw to order. Meanwhile, her piano playing is less precise and she cannot communicate the names of the pieces she plays. She has completely abandoned book reading and she looks only at the images of the news, in both newspapers and on televised news, to try and understand the headlines.

In July 2017, the patient had a consult at the Neuropsychology Unit of the Hospital Interzonal General de Agudos "Eva Perón" (*Eva Peron Interzonal General Hospital for Acute Diseases*) in San Martín, Buenos Aires, Argentina, for a suspected case of PPA and for supportive treatment of her increasing language difficulties, which were already hindering her everyday communications. She had begun to experience anomia and difficulties in understanding language seven years earlier, a year prior to retiring. Two previous neuropsychological evaluations had been administered in September 2014 and February 2017 (Table 1). A SPECT performed in October 2014 showed reduced blood flow in the following regions of the left hemisphere: the anterior basal temporal lobe, the medial frontal lobe and the anterior cingulate. A second SPECT, in March 2017, showed progression in the same compromised areas, with clearly reduced blood flow in the left anterior basal temporal lobe and slight to moderate reduction in blood flow in the medial frontal lobe and the anterior cingulate. An MRI performed in July 2017 showed cortical atrophy, predominantly of the left temporal lobe, affecting the temporal pole and the medial temporal lobe. Laboratory tests revealed only a mild hypercholesterolemia.

2.1.1.1 General neuropsychological evaluation. Patient BF was evaluated in 2014 and 2017 using a battery of tests that included the following: Clock Drawing Test (CDT; Freedman et al., 1994), Mini-Mental State Examination (MMSE; Folstein, Folstein, & McHugh, 1975, Spanish version from; Allegri et al., 1999), Spanish Boston Naming Test (SBNT; The Boston Naming Test, Kaplan, Goodglass, & Weintraub, 1983, Spanish version from, 1986; Allegri et al., 1997), Category and Letter Fluency Tasks from Spanish Verbal Fluency Test (Butman, Allegri, Harris, & Drake, 2000), Forward and Backward Digit Span subtest from III Wechsler Adult Intelligence Scale (WAIS III; Wechsler, 2002), Trail Making Test A and B (TMT; AITB, 1944; Reitan, 1958), Rey Complex Figure Test (RCFT; Meyers & Meyers, 1995), Design Fluency Test (DF): Five-Point Test (Lee, Strauss, McCloskey, Loring, & Drane, 1996; Regard, Strauss, & Knapp, 1982), California Verbal Learning Test (CVLT; Delis, Kramer, Kaplan, & Ober, 1987), Faux Pas Test (Stone, Baron-Cohen, & Knight, 1998; adult version for Buenos Aires from Zubizarreta Hospital) and the Reading the Mind in the Eyes Test (Eyes Test, Baron-Cohen, Jolliffe, Mortimore, & Robertson, 1997, adult version for Buenos Aires from Zubizarreta Hospital). Some of these tests were administered in just one of the two evaluations. The 2014 evaluation showed severe impairments in confrontation naming and in the category and letter fluency tasks. Her performance in the other evaluated cognitive domains was as expected for the patient's age and years of schooling.

Table 1. General neuropsychological evaluation.

Tests	09/09/14	02/08/2017
CDT	-	14/15
MMSE	-	27/30
		Z-scores
Language		
SBNT	−12.6	−39.8
Letter fluency	−2.3	−2.3
Category fluency	−2.5	−3.0
Attention		
Forward digits	−0.8	0.5
Backward digits	−1.1	−0.2
TMT A	-	−0.85
Visuoconstruction abilities		
RCFT Copy	-	0.1 (Copy Strategy II)
RCFT Time	-	1.27
Executive functions		
TMT B	-	−0.89
DF total unique designs	0.2	-
DF perseverations	0.5	-
CVLT–Memory		
Trial 1	-	−2.4
Learning	-	−2.8
List B	-	−0.9
Immediate Free Recall	-	−2.8
Immediate Cued Recall	-	−3.3
Delayed Free Recall	-	−1.6
Delayed Cued Recall	-	−1.7
Recognition	-	−0.5
Social cognition		
Faux Pas	0.1	-
Eyes Test	0.2	-

The second evaluation, carried out in February 2017, showed a worsened performance in the confrontation naming and categoric fluency tasks, as well as poor performance in the verbal memory evaluation and a pattern of deficits in information retrieval, though the patient's performance in this test may have been affected by her language impairment. Performance in other cognitive domains was spared, including: attention, visuoconstruction abilities, executive functions and social cognition (Table 1).

In conclusion, Patient BF was determined to have a prominent language impairment of a progressive nature that affected her performance in daily life. This profile is compatible with PPA, in accordance with Gorno-Tempini et al. (2011).

2.1.1.2 PPA classification. In July 2017, BF was evaluated using the BADA-R battery (abbreviated version, of the Battery for Analysis of Aphasic Deficits, Ferreres et al., 1999) in order to obtain a profile of the patient's language impairments. This battery allows the different components of language processing to be explored. The results showed an impairment of speech production due to severe difficulties in lexical retrieval (anomia). These difficulties were present both in the confrontation naming of objects and actions, as well as in in spontaneous speech, which was fluent, syntactically well-constructed and free of phonemic or phonetic problems but plagued by anomia and circumlocutions that tried to compensate for the lack of specific words. Slight impairments were also found in spelling to dictation, in which the patient produced phonologically plausible errors, and in the phonological short-term memory with a word span equal to four (Table 2).

Table 2. BADA-R results: BF's performance for the different tasks of the BADA-R. The number of errors in all tasks is reported, with the exception of the series of words repetition task, where the highest number of words that the patient can repeat immediately is reported.

Task	Results (errors/total)	Percentage of errors
Word repetition	0/15	0 %
Word reading	0/15	0 %
Word dictation	3/15	20%
Nonword repetition	0/18	0%
Nonword reading	0/18	0%
Nonword dictation	0/9	0%
Auditory lexical decision	0/24	0%
Visual lexical decision	1/24	4.16%
Auditory comprehension of objects	2/20	10%
Auditory comprehension of actions	0/20	0%
Visual comprehension of objects	2/20	10%
Visual comprehension of actions	0/20	0%
Oral naming of objects	13/16	81.25%
Oral naming of actions	6/16	37.5%
Sentence repetition	0/12	0%
Sentence reading	0/12	0%
Auditory comprehension of sentences	1/15	6.66%
Visual comprehension of sentences	1/15	6.66%
	Highest test achieved	
Series of words	4-WORD SERIES	

The auditory word-to-picture and written word-to-picture matching tests from the EPLA Battery (Kay, Lesser, Coltheart, Valle, & Vega, 1995; adapted version of the PALPA test by Kay, Lesser, & Coltheart, 1992) and the Pyramids and Palm Trees task (Howard & Patterson, 1992) were also administered in order to complete the semantic evaluation. BF exhibited a severely deficient performance in word-to-picture matching, with a Z-score of −11.04 and −11.39, respectively. In the Pyramids and Palm Trees task, the patient produced 20 errors (cut-off score: two errors).

The poor performance in naming, single-word auditory and visual comprehension, and semantic picture association suggests the presence of a semantic impairment.

The patient was also evaluated with the Spanish version of the Neuropsychological battery of the Uniform Data Set (UDS), FTLD Module (National Alzheimer's Coordinating Center & FTLD work group of the ADC Program, 2015. Spanish version from: Grupo de Trabajo Latino del Programa ADC y la Red Latinoamericana de Demencias, 2015), to identify whether she had one of the more frequent variants of PPA (Table 3).

The greatest difficulties in the performance of BF were observed in tasks aimed at evaluating semantic processing. Denomination and semantic association were the tests where more errors were committed. She failed to name any of the nouns and only achieved 25% success in the semantic association of animals. Her performance on the rest of the tests of the neuropsychological battery was very good (Table 3).

Considering the results of the applied neuropsychological batteries (BADA-R and UDS/FTLD) it is possible to answer the diagnostic key questions proposed by Marshall et al. (2018) in the clinical roadmap for diagnosis of canonical PPA variants. The patient BF has a prominent alteration of the language that affects her daily life. Within this alteration, it is possible to appreciate that her speech is not effortful or misarticulated, she does not commit frequent grammatical errors and she can repeat sentences correctly. Her difficulty

Table 3. UDS/FTLD module neuropsychological battery results.

Tests		Score	Percent correct
Semantic word-picture matching	Total correct	12/20	60%
Semantic Associates	Total correct	6/16	37%
	Animals	2/8	25%
	Tools	4/8	50%
Northwestern Anagram	Total correct	8/10	80%
	Relating to subject	5/5	100%
	Relating to object	3/5	60%
Sentence Repetition	Total correct	5/5	100%
	Words Omitted	0/37	–
	Semantic errors	0/20	–
	Phonological/other errors	0/20	–
Noun and Verb Naming	Nouns	0/16	0%
	Verbs	8/16	50%
Sentence Reading	Total correct	5/5	100%
	Words omitted	0/37	–
	Semantic errors	0/20	–
	Phonological/other errors	0/20	–

is focused on understanding the meaning of words. Brain imaging showed both atrophy and reduced blood flow to a temporal predominance, therefore, it is ruled out that the alteration of the language is due to other reasons (such as brain tumours or other non-degenerative pathologies). Following the diagnostic roadmap, these data lead us to state that BF can be identified as a case of svPPA.

In summary, taking into consideration the clinical history data, neuroimaging data results, neuropsychological assessments, language evaluations (BADA-R and UDS/FTLD, Tables 2 and 3) and the diagnostic roadmap of Marshall et al. (2018), clinical neurologists of the Neuropsychology Unit of the Hospital Interzonal General de Agudos "Eva Perón" established that Patient BF presents symptoms of svPPA in accordance with the consensus criteria proposed by Gorno-Tempini et al. (2011).

2.1.2 Control group

Since the tests used do not have local normative data, we decided to include a control group (CG).

The CG was comprised of seven adult women from the city of Buenos Aires and its suburbs, matched with the patient in terms of age ($M = 69.6$, standard deviation (SD) = 4.3; age range 64–75 years; $t = 0.305$, $p = .770$, $Z_{cc} = 0.326$) and schooling ($M = 15.9$, $SD = 2.7$; range 12–20 years; $t = -1.005$, $p = .354$, $Z_{cc} = -1.074$). The variable took into consideration the years of schooling that were successfully completed in the Argentine education system.

The inclusion criteria for admission to the CG were: to be women, native Spanish speakers, between 64 and 76 years of age, with a full secondary education and no neurological or psychiatric history. As the patient completed 13 years of schooling, dedicated her career to being a teacher and, in addition, was an avid reader (as stated by herself and her family) prior to the emergence of the first symptoms of her pathology, the inclusion criteria for schooling required that the subjects had completed secondary education and, at most, had led a professional career.

The participants were recruited from relatives and/or friends of the patients and professionals of the Neuropsychology Unit of the Hospital Interzonal General de

Agudos "Eva Perón", in San Martín, Buenos Aires, Argentina. A semi-structured interview was conducted to identify whether the applicants met the inclusion criteria.

2.2 Materials and method

Tests were administered to characterize the reading and writing impairments of BF, using spelling to dictation of regular and irregular words and nonwords, reading of words and nonwords and lexical decision with PSH.

The patient and controls were evaluated using the same set of tests, in the same order, after signing the informed consent. Tests were administered in two sessions of approximately 45 minutes each.

2.2.1 Dictation of regular and irregular words

A dictation test was designed in which regularity was controlled. The list consists of a total of 40 di- and trisyllabic stimuli: 20 regular words that are completely consistent, for example: ALTA (*tall*), MEDIDA (*measure*); and 20 irregular words, which are arbitrarily spelled words of varying degrees of inconsistency, for example: SAPO (*toad*), BUZÓN (*mailbox*). Both groups of words were balanced in terms of frequency (taken from the *BuscaPalabras* database by Davis & Perea, 2005) and length, and they were presented in a pseudorandom order. The evaluator dictated the words to the participants, who had to write them down on a blank page. The participant chose the allograph they wished to use, as well as the distribution of the words on the page.

2.2.2 Dictation of nonwords

A task was prepared that included the dictation of 20 nonwords (pseudowords) formed from the recombination of the syllables of 10 regular words and 10 irregular words, which were taken from the spelling to dictation task described above. The selected words were balanced in terms of frequency (five high-frequency words and five low-frequency words) and length (five disyllabic words and five trisyllabic words). The recombination of syllables respected the position of the syllables in the base word. The evaluation procedure followed the same method as was used in the regular and irregular spelling to dictation test.

2.2.3 Reading of words and nonwords from the transpruebas battery (China & Ferreres, 2017)

The Transpruebas Battery allows the lexical and sublexical processes involved in the naming, repetition, reading, and dictation tasks to be studied. In this present investigation, we only used the reading aloud of words and nonwords test. It consists of a total of 90 stimuli (45 words and 45 nonwords). The words consist of 45 concrete drawable nouns of two, three and four syllables, which are controlled for length, frequency and syllabic complexity. The 45 nonwords consist of recombinations of the syllables of the words described above and are controlled in terms of length, accentual contour, syllabic complexity, frequency of syllables and phonemes.

The stimuli were presented on the screen of a laptop using the *SuperLab* software (Beringer, 1995). Participants were asked to read the stimuli as quickly and accurately as possible. Response latencies were recorded by a voice key connected to the computer.

2.2.4 Lexical decision with pseudohomophones (Difalcis, Leiva, Ferreres, & Abusamra, 2018)

The test consists of 53 words, 53 nonwords and 53 PSH. The 53 words (Word List A) are high- and low-frequency nouns without accents. The list is matched in terms of both lexical variables (frequency and number of orthographic neighbors) and sublexical variables (length in letters, and type and token bigram frequency). Following the suggestion of Martin (1982), the 53 nonwords and 53 PSH were created by modifying a single letter from a different list of words (Word List B) than previously used, with the same variables controlled. For example, from the word "noticia" (*news*), the nonword "notifia" and the PSH "notisia" were created. Word List B is not included in the task, and therefore the subjects do not see the list of words from which the nonwords and PSH are derived. The test also includes 53 mid-frequency filler words to balance the number of words with the nonwords and PSH.

The stimuli were presented on the screen of a laptop using the *SuperLab* software (Beringer, 1995). Participants were asked to press the letter S on the keyboard if they though it was a word and N if they thought it was a nonword. RT and number of hits were recorded.

2.3 Data analysis

Data cleansing was performed prior to carrying out the RT analysis of the "Reading of words and nonwords" test in order to eliminate RT corresponding to erroneous responses or technical faults as well as to excessively short or long RT. 60/720 RT (8.3%) were eliminated from the analysis, as they corresponded to erroneous responses of participants and/or technical faults. Of the remaining 660 RT, 19 RT (2.9%) were eliminated from the analysis because they were very short or very long, with a cut-off of the ≤ 1st percentile and ≥ 98th percentile considered for each type of stimulus. Words with RT of < 409 ms and > 1188 ms and nonwords with RT of < 446 ms and > 2037 ms were eliminated from the control group. For the patient, words with RT of < 681 ms and > 5053 ms and nonwords with RT of < 488 ms and > 5292 ms were eliminated.

2.3.1 Analysis

First, the descriptive statistics of the number of hits for all tests and the RT in the reading test were calculated for the patient and the CG. The modified *t* test (Crawford & Howell, 1998), designed for single cases, was used to compare the patient's performance against that of the CG. To analyze the patterns of acquired dyslexia and dysgraphia in the patient, the presence of dissociations in hits and RT (if applicable) were sought between: 1) writing to dictation: regular vs. irregular words and irregular words vs. nonwords; 2) reading of words and nonwords (RT); 3) lexical decision: words vs. nonwords and PSH vs. nonwords. We used the operational criteria for classical and strong dissociation by Crawford and Garthwaite (2005). A classical dissociation is assumed when the following criteria are met: 1) the patient scores significantly lower than the CG ($p < .05$) in task A; 2) the patient does not have a difference in performance from the CG ($p > .05$) in task B; and 3) the observed difference between tasks A and B in the patient are statistically different ($p < .05$) than the difference observed in the CG on the same tasks. In the case of strong dissociations, criterion 2 changes, as it states that the patient's performance may show statistically significant differences from the CG. The modified *t* test (Crawford & Howell, 1998) was used to analyze compliance with criteria 1 and 2, and for criterion 3, the *Revised Standardized Difference Test* (RSDT) was used, which compares the discrepancy between tasks in the patient and the CG, taking into account the correlation between them (Crawford & Garthwaite, 2005). The percentage of the healthy population that would score a lower performance than the patient was also estimated using the Crawford and Garthwaite (2005) method, and the size of the effect (z_{cc}) is reported for all comparisons (Crawford, Garthwaite, & Porter, 2010).

In addition, the Chi^2 test was used to compare the difference between hits and errors in the patient for: 1) writing to dictation of: regular vs. irregular words and irregular words vs. nonwords; 2) reading of words vs. nonwords; 3) lexical decision (PSH vs. nonwords). As for the reading times of the patient, the *t* test was used for related samples to evaluate the difference in RT between words and nonwords.

Finally, the CG was evaluated for the presence of a lexical advantage in reading and the pseudohomophone effect. For the former, the *t* test was used for related samples (RT of words and nonwords). For the latter, given that the distribution did not conform to a normal curve, the Wilcoxon signed-rank test was used (number of hits for PSH vs. nonwords).

3. Results

Table 4 shows the scores achieved by the patient in each of the experimental tests, and the comparison analysis between tests and between types of stimuli in each test. It also shows the performances of the CG for the same tests and the results of the comparison between the patient and the CG.

3.1 Writing

On the dictation tests, Patient BF scored a very low performance on irregular words (4/20 correct, 20%), which contrasts with her optimal performance on regular words (20/20, 100%) and nonwords (20/20, 100%). The difference between irregular and regular words was statistically significant (Chi^2 = 26.667, $df = 1$, $p < .001$), which indicates the presence of a regularity effect. Likewise, the difference between irregular words and nonwords was significant (Chi^2 = 26.667, $df = 1$, $p < .001$). When compared with the control group, BF only had statistically significant differences for the writing of irregular words, but not for regular or nonwords (Table 4). The error type analysis revealed that BF only produced errors with irregular words, and almost all of these were phonologically plausible errors (15/16, 93.75%), for example: JUVENTUD > JUBENTUD (*youth*), SITIO > CITIO (*site*), etc. (see Appendix A). At the same time, the RSDT analysis showed that the difference between regular and irregular words observed in BF was significantly greater than that observed in the control group, and with a very low probability of observing such a discrepancy in the population (less than 1%) (Table 5). This result indicates that the patient meets the criteria for a classical dissociation between the spelling to dictation of regular and irregular words, with a low performance only in the latter. Furthermore, BF presented a significantly greater difference between irregular words and nonwords than was observed in the control group, and the probability that this could be observed in the population was

Table 4. Scores achieved by the patient in the experimental tests, performances of the CG and comparison between BF and CG.

		Patient BF		CG (n = 7)	BF vs CG[d]		
Tests	*Stimuli*	*Correct/total or M (SD) of Rt*[a]	*Intrasubject comparison (p)*	*M (SD)*	*t*	*p*	*Estimated effect size (z_{cc})*[e] *Point (95% CI)*
Dictation	Regular words	20/20	<.001 b ***	19.7 (0.8)	0.351	.369	0.4 (−0.4 to 1.1)
	Irregular words	4/20	<.001 b ***	19.6 (0.8)	−18.241	<.001***	−19.5 (−30.3 to −8.8)
	Nonwords	20/20		19.1 (0.7)	1.166	.144	1.2 (0.2 a 2.2)
Reading: Accuracy	Words	45/45	.153 b	44.9 (0.4)	0.234	.411	0.3 (−0.5 to 1.0)
	Nonwords	43/45		43.4 (1.3)	−0.288	.391	−0.3 (−1.1 to 0.5)
Reading: latencies	Words	1352.6 (318)	.579 c	730.6 (78.5)	7412	<.001***	7.9 (3.5 to 12.3)
	Nonwords	1438.6 (883.3)		918.7 (194.9)	2495	.002**	2.7 (1.0 to 4.3)
Lexical Decision	Words	44/53		52.6 (0.5)	−16.089	<.001***	−17.2 (−26.7 to −7.7)
	Nonwords	52/53	<.001 b ***	52.6 (0.5)	−1.122	.153	−1.2 (−2.2 to −0.2)
	Pseudohomophones	38/53		51.6 (1.0)	−12.722	<.001***	−13.6 (−21.1 to −6.1)

a RT = Reaction time: higher scores indicate worse performance. b Chi2 test. c independent t test. d Crawford and Howell (1998), the results are for a one-tailed test. e Crawford et al. (2010). Level of statistical significance: * = $p < .05$; ** = $p < .01$; *** = $p < .001$. M: media; (SD): standard deviation; CG: Control Group

Table 5. Results of the revised standarized differences test (RSDT) for the comparisons of the differences between tests and estimated percentage of the healthy population that would present a greater discrepancy than the observed in patient BF.

Comparisons	Correlation between tasks in CG	RSDT patient vs CG [a]		Estimated % of the healthy population with a greater discrepancy than BF [b]	Dissociation
		t	*p*		
Dictation					
Irregular words vs regular words	r = .509	10.650	<.001***	<1%	Classical
Irregular words vs nonwords	r = 547	11.178	<.001***	<1%	Classical
Reading					
Words vs nonwords	r = .468	0.443	.673	34%	None
Reading: RT					
Words vs nonwords	r = .666	4.556	.003**	<1%	Strong
Lexical decisión					
Pseudohomophones vs nonwords	r = −.454	6.238	<.001***	<1%	Classical

[a]Crawford and Garthwaite (2005) [b] Crawford & Garthwaite (2002). Level of statistical significance: *p < .05; **p < .01; ***p < .001

estimated to be less than 1% (Table 5). This indicates that the patient also meets the criteria for a classical dissociation between irregular words and nonwords, with impaired performance only for the former, as mentioned above.

3.2 Reading

Patient BF's reading accuracy was very good, both in terms of words and nonwords, with no significant difference between the types of stimuli ($Chi^2 = 2.045$, $df = 1$, $p = .153$), nor a significant difference with the CG (Table 4).

On the other hand, in the reading RT, BF showed prolonged latencies and the loss of lexical advantage. BF's latencies were significantly longer than those of the CG, both in words and nonwords (Table 4). The prolonged RT were relatively longer for the reading of words (7.9 SD above the mean of the CG) than nonwords (2.7 SD above the mean of the CG), which implies a decrease in the difference between the RT of both stimuli, that is to say, a decrease/disappearance of the lexical advantage in the RT. Indeed, the intrasubject analysis showed there were no significant differences in the RT of both types of stimuli (t words vs. nonwords $_{BF} = -0.559$, $gl = 47.911$, $p = .579$). The lexical advantage was present in the CG (t words vs. nonwords $_{CG} = -3.228$, gl = 6, p = .018). Consistent with this result, the RSTD analysis showed that the small difference observed in BF between the RT of words and nonwords (lack of lexical advantage) was significantly different from that observed in the CG (presence of lexical advantage), which indicates the presence of a strong dissociation (Table 5).

3.3 Lexical decision with pseudohomophones

In the lexical decision test, the stimuli in which Patient BF produced the greatest number of errors (false positives) were the PSH, where she scored 38/53 correct responses (72% correct hits). The patient also committed errors (false negatives) with words (44/53 correct, 83%) and scored an almost perfect performance with nonwords (52/53, 98%). The comparison between the different types of stimuli evidenced the pseudohomophone effect, that is, a significantly lower performance in PSH than in nonwords ($Chi^2 = 14.428$, $df = 1$, $p < .001$), an effect that was not recorded in the CG ($Z = -1.633$, $p = .102$). BF also scored a lower performance in words than nonwords ($Chi^2 = 7.067$, $df = 1$, $p = .008$) compared to the CG, which scored an equal performance in words as in nonwords (Table 4). In the comparison between BF and the CG, significant differences were observed in PSH and words (Table 4). The RSDT analysis of the dissociations in performance between the different types of stimuli showed that the patient met the criteria for a classical dissociation for the comparison between PSH and nonwords, and also for the comparison between words and nonwords (Table 5).

4. Discussion

The aim of this paper was to report the writing and reading impairments of BF, a Spanish-speaking patient with svPPA, and show that it is possible to identify patterns of acquired surface dysgraphia and dyslexia in a transparent language, such as Spanish, when the linguistic particularities are taken into consideration in the selection of tests. To do so, a dictation test was designed and reading tests that had already been used in the local setting and that control the relevant variables were administered. In addition, an intra-subject analysis and a comparison analysis with a control group were carried out to test the dissociations that characterize these patterns.

BF's test results for spelling to dictation showed a clear regularity effect, which implies the correct writing of regular words and nonwords, and a clear deterioration in the writing of irregular words, stimuli with which phonologically plausible errors were produced. These results would be consistent with those found in patients with focal lesions in whom acquired surface dysgraphia was identified (Ferreres et al., 2012, 2005; García Orza et al., 2002; Iribarren et al., 2001). Furthermore, the RSDT analysis clearly showed classical dissociations between regular and irregular words and between nonwords and irregular words, with impairment only of the irregular words in both cases, providing evidence of the regularity effect in Patient BF's spelling to dictation.

Some studies of Spanish-speaking patients have objected to the value of phonologically plausible errors for the identification of acquired surface dysgraphia (Ardila, 1998; Ardila, Rosselli, & Ostrosky-Solis, 1996). Arguments were based on the fact that these errors – which have also been called homophone or orthographic errors by other authors (Ardila, 1998)[3] – tend to be found in the healthy population, particularly in participants with low levels of schooling. However, we believe that this objection does not apply in this case, for two reasons. First, Patient BF has a high level of education and was professionally active in the teaching field and, thus, arguably has good reading and writing skills. And second, the CG, matched in gender, age and years of schooling with the patient, scored close to maximum performance in the spelling to dictation of irregular words (Table 4).

Thus, the spelling to dictation results support the argument that Patient BF shows an acquired impairment in the writing of irregular words, wherein she produces phonologically plausible errors that cannot be attributed to low schooling and shows preservation of the dictation of regular words and nonwords. This performance is compatible with acquired surface dysgraphia and, within the framework of the dual route models, it can be interpreted as an impairment of the lexical route of writing with preservation of the non-lexical route.

This case study shows that it is possible, even relatively simple, to administer tests that also allow the regularity effect to be found in Spanish writing. This indicates that it is not necessary to forgo the identification of acquired surface dysgraphia as part of the criteria for the diagnosis of svPPA in Spanish-speaking patients. The existence of a significant number of irregular words for writing in Spanish makes it entirely possible to evidence the characteristic dissociation of the pattern of acquired surface dysgraphia. As is the case in opaque writing systems, and as proposed by the international diagnostic criteria (Gorno-Tempini et al., 2011), all that is necessary is to use lists of regular and irregular words and nonwords in the dictation, and an appropriate CG. Naturally, and according to the work of Ardila et al. (1996) and Ardila, Matute, and Inozemtseva (2003), the case can also be interpreted as a "regression" that affects irregular words. According to these authors, writing irregular words is more complex than writing regular words, and is achieved later in the learning process. Thus, the pathology produces a regression in the order of acquisition that affects the most difficult stimuli first. Although this could be the case for acquired surface dysgraphia, this hypothesis does not explain two patterns that have already been documented in Spanish: acquired phonological dysgraphia (Iribarren et al., 2001) and acquired deep dysgraphia (Davies & Cuetos, 2005). These two profiles present with relative spared orthographic knowledge (more difficult) with a severe deterioration of writing through phoneme-to-grapheme conversion (easier). The dual route model of writing provides an explanation for all of these cases and predicts the finding of different patterns of acquired dysgraphia in different types of PPA, as has already been documented in English-speaking patients (Neophytou, Wiley, Rapp, & Tsapkini, 2019).

With regard to the identification of the pattern of acquired surface dyslexia in a transparent writing system such as Spanish, the strategy we adopted for Patient BF consisted of: a) trying to demonstrate, through the measurement of reading latencies, that BF used the same non-lexical route for reading words and nonwords; and b) demonstrating the loss of orthographic information through a visual lexical decision with PSH test, as was used in the study of patients with focal lesions (Ferreres et al., 2012, 2005).

The good accuracy shown by BF in the reading of words and nonwords, and the lack of significant differences between both types of stimuli in terms of number of hits, is compatible with the use of a non-lexical route for reading since, as pointed out in the introduction, all Spanish words can be read correctly if the mechanism for reading through the application of grapheme-to-phoneme rules is preserved. However, this result does not constitute direct evidence that the lexical reading route has been impaired. The lack of difference in accuracy in the reading of words and nonwords (by Spanish speakers) is a necessary sign, but it is not enough to conclude there is a pattern of acquired surface dyslexia. The reading latencies of both words and nonwords must be taken into consideration. In this case, BF did differ from the controls: while the CG showed lexical superiority (shorter RT for words than for nonwords), a loss of lexical advantage was observed in BF and RT were prolonged but virtually equal for both types of stimuli. The RSTD analysis confirmed that the latencies observed in BF were significantly different from those of the control group, which constitutes a strong dissociation. The equalization of word and nonword processing times has already been interpreted as the result of the impairment of the lexical reading route in Spanish-speaking patients with acquired surface dyslexia (Ferreres et al., 2012, 2005; Valle-Arroyo, 1996). However, from a clinical perspective, this measurement is cumbersome because it requires devices that are not usually found in the clinical environment, such as a voice key and the software needed to measure RT. In the future, it would be appropriate to make ecological adaptations to the task, such as, for example, an analysis to detect relevant stimuli so as to shorten the list of items, or an evaluation of other simpler measures of timing, such as the total time per stimulus type, among other adaptations.

As for the result of the lexical decision with PSH, the finding of PSH disadvantage (greater difficulties with PSH than with nonwords) is evidence that BF cannot rely on orthographic information to decide if a PSH is a legal orthographic word. It is our interpretation that, in lacking the orthographic information, BF relies on phonological mediation and makes lexical decisions through consulting her phonological lexicon. Since phonological mediation applied to a PSH activates the representation of a phonological word, the patient commits the false positive error of believing the PSH is a word. The pseudohomophone effect was not observed in the CG and, in BF, the RSDT analysis confirmed a classical dissociation between PSH and nonwords. These results indicate that the lexical decision test is useful for detecting lexical reading impairments and, specifically, an impairment of the orthographic input lexicon.

5. Conclusion

In conclusion, this case study shows that: a) it was possible to identify the pattern of acquired surface dysgraphia and dyslexia in a Spanish-speaking patient with svPPA; b) the identification of acquired surface dysgraphia does not require the use of different measurements than those used in opaque writing systems; c) two of the tasks tested in patients with focal lesions can be used for the identification of acquired surface dyslexia in patients with svPPA: the lexical decision with PSH and the measurement of word and nonword reading latency tasks; and d) considering the results found, it is not necessary to forgo the testing of the criteria for the possible presence of acquired surface dyslexia or dysgraphia for the diagnosis of svPPA in Spanish-speaking patients.

Notes

1. In Rioplatense Spanish.
2. Another rule states that when the H is preceded by the letter C, it forms the grapheme CH, which is pronounced /ĉ/ as in CHANCHO > / ĉanĉo/ (*pig*).
3. Phonologically plausible, orthographic and homophone errors refer to the same phenomenon: errors in writing the correct sequence of words, though the result "sounds" the same as the correct word.

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Appendix

Patient BF spelling to dictation of irregular words.

Blank stimulus	English	Response
Sastre	Tailor	Sastre
Precio	Price	Precio
Juventud	Youth	Jubentud
Buzón	Mailbox	Busón
Higuera	Fig tree	Higera
Sitio	Site	Citio
Sapo	Toad	Zapo
Posición	Position	Posicion
Escena	Scene	Escena
Origen	Origin	Horigen
Omisión	Omission	Homicion
Joya	Jewel	Jolla
Pulsera	Bracelet	Pulsera
Vocablo	Word	Bocablo
Yeso	Cast	Yezo
Zona	Zone	Sona
Esbozo	Sketch	Ezboso
Nivel	Level	Nibel
Belleza	Beauty	Velleza
Plaza	Square	Plasa