

Preserved visual language identification despite severe alexia

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ABSTRACT

Patients with letter-by-letter alexia may have residual access to lexical or semantic representations of words despite severely impaired overt word recognition (reading). Here, we report a multilingual patient with severe letter-by-letter alexia who rapidly identified the language of written words and sentences in French and English while he had great difficulty in reading them, judging their lexical status or extracting semantic information. Lexical decision was strongly influenced by the orthographic structure of stimuli: whereas he easily determined the lexical status of illegal nonwords (e.g., 'rsdo'), he had random performance with legal pseudowords (e.g., 'binus'). When asked to determine the language of meaningless letter trigrams with high frequency in the English or French orthography (e.g., 'oth' or 'iqu') his performance was significantly above chance. In contrast, similarly to healthy participants his language decision was at chance with low-frequency trigrams. These findings suggest that written language identification relies on sublexical processing of orthographic rules specific to each language.

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1. Introduction

Letter-by-letter alexia (LBL) is an acquired reading disorder consecutive to left occipito-temporal damage centered on the fusiform gyrus (Cohen et al., 2003; Damasio & Damasio, 1983). Though naming of isolated letters is often preserved in patients with LBL alexia word reading is laborious and characterized by a letter-by-letter identification strategy. As a result, the time to identify a word is linearly related to the number of its constituting letters, with an increase of up to 2 s or more for each additional letter (Behrmann, Nelson, & Sekuler, 1998; Leff et al., 2001). Current accounts posit that the impairment preventing patients with LBL reading from adopting the parallel process observed in normal readers affects peripheral processes such as the visuo-orthographic level (Arguin & Bub, 2005; Behrmann, Plaut, & Nelson, 1998; Patterson & Kay, 1982).

In spite of their laborious reading some LBL readers are able to categorize words according to their semantic and/or lexical status, suggesting at least partial activation of higher-level lexical-semantic representations (Behrmann, Plaut, et al., 1998; Lambon Ralph, Hesketh, & Sage, 2004). Evidence for this phenomenon – termed the Saffran effect – is the fact that some LBL readers perform well above chance in lexical or in semantic decision tasks (Coslett, Saffran, Greenbaum, & Schwartz, 1993; Landis, Regard, &

Serrat, 1980; Saffran & Coslett, 1998). Here, we present a LBL patient (AL) who demonstrates a form of Saffran effect that has not been described yet: he was able to identify rapidly and without effort the language of words and sentences that he failed to read. Several neuropsychological and neurophysiological studies support the idea that the human brain, as a result of perceptual experience, encodes the orthographic structure of a specific language (McClelland & Rumelhart, 1981). More precisely, Binder et al. (Binder, Medler, Westbury, Liebenthal, & Buchanan, 2006; see also Dehaene, Cohen, Sigman, & Vinckier, 2005) demonstrated that the left lateral fusiform gyrus, a region whose damage is critical for the occurrence of pure alexia, is sensitive to the familiarity of letter combinations. According to these findings, we hypothesize that AL's language decision might be based on the detection of local orthographic regularities that are specific to each language. For example, the letter trigram 'oth' is common in English, but does not exist in French whereas the trigram 'iqu' is frequent in French, but not in English. Based on this hypothesis we expected that AL is capable of identifying the language of nonsense letter combinations provided they have a high frequency of occurrence in a specific language.

2. Material & Methods

2.1. Patient description

AL was a highly educated, 72-year-old right-handed man who suffered a left medial occipito-temporal ischemic stroke (Fig. 1) causing pure alexia and right superior quadrantanopia. He was a native French-speaker. He had excellent knowledge of English and

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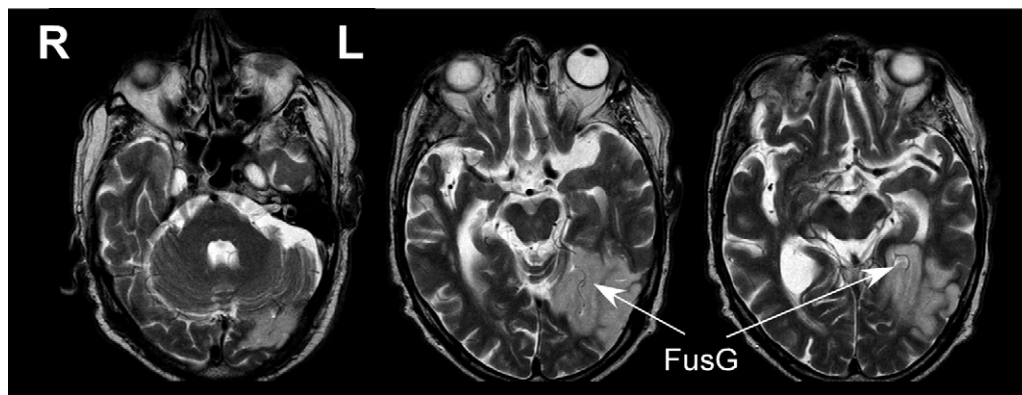


Fig. 1. T2-weighted MRI-scan showing ischemic damage to the left occipito-temporal cortex involving the fusiform (FusG), lingual, lateral occipital, and posterior parahippocampal gyrus.

Table 1
Results of neuropsychological evaluation.

Function	Test	Patient raw score (%)	Percentile	Classification
Naming on definition	Boston Naming Test (1)	19/20 (95%)	>50	High average
Repetition	Clinical protocol	44/44 (100%)		High average
Oral comprehension	MT-86 (2)	43/47	–	High average
Writing (oral spelling)	Items from de Partz (3)	Regular words 19/20 Irregular words 18/20	– –	Average Average
Reading from oral spelling	Examen des Dyslexies Acquis (4)	97/99 (98%)	–	High average
Visual recognition	VOSP incomplete letters	14	18.8	Impaired
	VOSP Object Decision	16	17.7	Low average
	VOSP Dot Counting	10	9.9	High average
	VOSP Position Discrimination (5)	20	19.6	High average
Memory	Warrington words (6)	43	43	High average
Executive functions	Verbal fluency (7)	34	34.2	Average
	Figural fluency (8)	16	26.3	Low average

1. Kaplan, Goodglass, & Weintraub (1983); 2. Nespoulous et al. (1992); 3. de Partz (1994); 4. Lemay (1990); 5. Warrington and James (1991); 6. Warrington (1984); 7. Thurstone and Thurstone (1963); 8. Regard (1981).

good knowledge of German (languages he had often used during his professional life), though he had mostly stopped to use these languages since the time of his retirement (seven years prior to this study). Using the classification of Roux et al. (2004) at the time of this study AL would be classified as LIII (acquisition of English and German after the age of 7 years, not spoken every day). Standardized neuropsychological testing revealed moderate deficits of verbal and non-verbal memory, executive functions and visual identification (Table 1). Other cognitive functions, including ideomotor praxis, arithmetic skills and short-term memory, were normal. Oral production of language, comprehension and repetition were normal, and writing was intact. The most apparent deficit was an inability to read visually presented material, while identification of words orally spelled was virtually perfect. He showed extremely laborious letter-by-letter reading, sometimes confusing even single letters, and was often unable to identify a word even under unlimited exposure. For example, AL would identify the word “étoile” (star) as /e/ /tO/ /i/ /b°/ /e/ (corresponding to: é-to-i-beé) in a first attempt, and subsequently as /etwab/ (corresponding to: étoibe) which took him more than 10 s. Amazingly, when asked to indicate in what language a text that he was unable to read was written (English, French, or German), he immediately recognized the language. Given the absence of aphasia in French, and the fatigability of the patient, we decided not to examine spoken English.

2.2. General methods

The study started 3 months after the stroke and lasted for 6 months. During this period the patient's reading deficit remained

stable. Evaluation of reading was performed in French and English. The patient's performance was compared to three age- and gender-matched healthy volunteers (3 males, mean age, 71 ± 2.7 years). All were native French speakers with excellent knowledge of written English which had been acquired after the age of 7 years and not spoken every day (proficiency classified as LIII according to Roux et al. (2004)). The study was approved by the Ethics Committee of the University Hospital of Geneva, and all participants gave informed, written consent.

Before every experiment, several practice trials were performed. Item presentation in each experiment was pseudo-randomized and counterbalanced across control-subjects. Stimuli were presented centrally on a 17" computer-screen in black letters (Arial, size 36) on white background. A central fixation cross was presented for 1000 ms, followed by a blank screen (100 ms), and the stimulus (either 400 ms or unlimited, as specified below). In the naming and reading tasks, responses were registered with a voice-sensitive key with millisecond accuracy. In the forced-choice tasks, participants responded by pressing one of two keys on a button box with their left and right index finger.

2.3. Reading

2.3.1. Methods

Reading of letters was evaluated by asking participants to name each letter of the Latin alphabet presented for unlimited time. Upper and lowercase letters were presented in pseudo-random

Table 2A
Summary of reading performance.

	Exposure time	Errors raw score (%)	Total error rate (%)	Normal values (mean errors raw score 3 controls)	p^*
Letter reading					
Uppercase	∞	7/52 (13%)	14%	0/52 (0%)	
Lowercase	∞	8/52 (15%)		0/52 (0%)	
Word reading					
French, high-frequency	∞	6/17 (35%)	46%	0/17	$\chi^2 = 1.44$, ns
French, low-frequency	∞	11/20 (55%)		0/20	
English, high-frequency	∞	11/24 (46%)	53%	0/24	$\chi^2 = 0.92$, ns
English, low-frequency	∞	16/27 (59%)		0/27	
French, high-frequency	400 ms	13/17 (77%)	81%	nt	$\chi^2 = 0.44$, ns
French, low-frequency	400 ms	17/20 (85%)		nt	
English, high-frequency	400 ms	20/24 (83%)	88%	nt	$\chi^2 = 1.05$, ns
English, low-frequency	400 ms	25/27 (93%)		nt	

ns, not significant.

* p : comparison between high- and low-frequency words for unlimited and 400 ms exposure time.

order in a single block of 52 stimuli. Two trials were made on two separate sessions. Reading accuracy and latency was assessed.

Word reading was assessed by asking participants to read aloud French and English words. The French list contained 37 words of high ($n = 17$) and low frequency ($n = 20$) taken from the EDA Battery (Lemay, 1990). Word length was 3 ($n = 5$), 4 ($n = 6$), 5 ($n = 6$), 6 ($n = 7$), 7 ($n = 8$), 8 ($n = 4$) and 9 ($n = 1$) letters. The English list contained 51 words of high ($n = 24$) and low frequency ($n = 27$) taken from the PALPA Battery (Kay, Lesser, & Coltheart, 1992). Word length was 3 ($n = 1$), 4 ($n = 11$), 5 ($n = 12$), 6 ($n = 12$), 7 ($n = 11$), 8 ($n = 2$) and 9 ($n = 2$) letters. The distribution of words as a function of word length and frequency is shown in Table 2B. French and English words were presented in the same block and in pseudo-random order.

2.3.2. Results

Table 2A shows the patient's error rate in letter and word reading. Reading was extremely laborious with a difficulty to identify single letters when embedded within words or when presented alone (14% errors, 15/104). AL's difficulties with single letter reading did not differ significantly from his performance when identifying fragmented letters of the Visual Object and Space Perception Battery (Warrington & James, 1991) ($\chi^2 = 2.67$, ns), suggesting that degrading perceptual information did not exacerbate his reading deficit.

When there was no time limitation, AL failed to read a comparable proportion of French and English words (46% and 53%, respectively; $\chi^2 = 1.44$, ns). Most reading errors were neologisms: for example, AL read 'gazon' (French 'lawn') as 'dizal' and 'cart' as 'rast'. When exposure was limited to 400 ms, a presentation time at which healthy readers easily recognize single words, he failed to read 81% French and 88% English words ($\chi^2 = .87$, ns). In order to investigate more precisely the effect of exposure time, we analyzed AL's reading errors separately for French and English, high- and low-frequency words. Not surprisingly, the patient performed better with unlimited presentation time as compared with 400 ms both for high- and low-frequency words (French high-frequency words: $\chi^2 = 5.85$, $p < .05$; French low-frequency words: $\chi^2 = 4.29$, $p < .05$; English high-frequency words: $\chi^2 = 7.38$, $p < .01$; English low-frequency words: $\chi^2 = 8.21$, $p < .01$). More importantly, AL's reading was not affected by word frequency. His performance was comparable for words of high- and low-frequency, whether we examined the entire list (Table 2A) or tested for frequency effects as a function of word length (Table 2B).

One of the hallmarks of LBL is a linear increase of reading time with increasing word length; this is generally evaluated with simple regression. Unfortunately, due to his severe alexia AL read only 20 French and 24 English words correctly under unlimited exposure time. This number further decreased following exclusion of four outliers (reading time > 15 s). Nevertheless, a normality test

(Kolmogorov–Smirnov) showed that the distribution of reading times was normal. Even with this limited data set we found that AL's reading times linearly increased as a function of word length (French and English combined: $F(1,39) = 4.93$, $p < 0.5$; linear trend: 1040 ms/letter; Fig. 2), confirming the serial, letter-by-letter reading strategy typical for pure alexia. In contrast, the reading speed of the control participants was independent of word length (linear trend: 8 ms/letter; not significant).

In sum, AL's reading performance was comparable for French and English, independent of word frequency, but strongly affected by word length. These characteristics indicate that the patient suffered from a peripheral reading disorder and justify a classification of his disorder as pure alexia.

2.4. Lexical and semantic decision

2.4.1. Methods

The severe impairment of letter and word identification and the effect of word length indicated that AL had pure alexia. Some LBL readers may extract lexical or semantic information from words that they are unable to identify explicitly (Coslett & Saffran, 1989; Landis et al., 1980). Access to lexical or semantic knowledge would provide an explanation of AL's ability to identify the language of written material. We therefore next examined whether our patient was able to extract lexical and semantic information from French and English words.

In the lexical decision task, participants were instructed to indicate whether a written stimulus (word or pseudoword) represented an existing word or not. The French corpus contained 96 stimuli (EDA battery; Lemay, 1990), half being real words (word length: 3–9 letters) of high ($n = 25$) or low frequency ($n = 23$) and the other half pseudowords (36 orthographically plausible, like 'chopadart'; 12 orthographically non-plausible, like 'rlecpaefe'). The English corpus contained 180 words (PALPA Battery; Kay et al., 1992) half being real words (word length: 3–9 letters) of high ($n = 45$) or low frequency ($n = 45$) and the other half pseudowords (60 orthographically plausible, like 'binus'; 30 orthographically non-plausible, like 'rsdo').

The semantic decision task tested whether AL was able to extract categorical information from visually presented words. The task was to classify words according to whether they belonged to the category of fruits or not. For 400 ms exposure time, 36 high-frequency French words and the same number of English words, belonging to one of four semantic categories (fruit, animal, furniture or transport) were selected (word length: 4–10 letters). For 1000 ms exposure time, the experiment was performed with 32 French and 32 English words. Testing was performed separately for in French and English.

Table 2B
Reading performance under unlimited time across word-letter lengths and frequency.

		3–4 letters	<i>p</i>	5–6 letters	<i>p</i>	7–8–9 letters	<i>p</i>
English	Low frequency	5/8 (.63)	$\chi^2 = 0.18$, <i>p</i> ns	9/13 (.69)	$\chi^2 = 2.59$, <i>p</i> ns	2/6 (.33)	$\chi^2 = 0.18$, <i>p</i> ns
	High frequency	3/4 (.75)		4/11 (.36)		4/9 (.44)	
French	Low frequency	5/6 (.83)	$\chi^2 = 0.74$, <i>p</i> ns	4/6 (.67)	$\chi^2 = 1.88$, <i>p</i> ns	2/8 (.25)	$\chi^2 = 0.04$, <i>p</i> ns
	High frequency	3/5 (.60)		2/7 (.29)		1/5 (.20)	

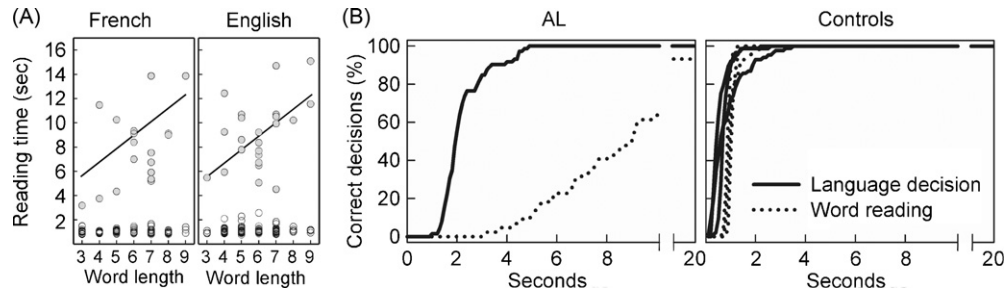


Fig. 2. (A) Reading times (only correct responses) for the patient (filled circles) and control participants (empty circles) as a function of word length. The diagonal line shows the linear trend. (B) Cumulative distribution of reaction times in the language decision and word reading tasks.

2.4.2. Results

2.4.2.1. Lexical decision. AL made 28/96 (29%) errors with French words. He classified 10/48 (21%) real words as non-existent words, and his performance with pseudowords was significantly influenced by orthographic plausibility. Indeed, lexical decision with plausible pseudowords (18/36 errors; 50%) was significantly worse than with non-plausible pseudowords (0/12 errors; $\chi^2 = 9.6$, $p = .001$). In English, overall performance was 68/180 errors (38%). AL made 21/90 (23%) errors with real words and produced a significantly higher error rate with plausible (45/60 errors, 75%) than with non-plausible pseudowords (2/30 errors, 7%; $\chi^2 = 37.42$, $p < .001$). The stronger tendency in English compared to French to accept plausible pseudowords as real words can be interpreted in terms of differences in language proficiency: AL might have had stronger control over his tendency to accept regular pseudowords in French because his lexicon was richer in this language.

Given the high frequency of lexical decision errors with plausible pseudowords in English, we compared AL's performance across languages. Comparison of overall performance in French and English was not significant ($\chi^2 = 2.05$, ns). In contrast, while AL had a stronger tendency in English than in French to classify plausible pseudowords as existing words ($\chi^2 = 6.23$, $p < 0.5$), no such difference was observed for non-plausible pseudowords ($\chi^2 = 0.84$, ns).

Healthy participants made on average 1.7 (2%) errors in French and 5.67 (5%) errors in English.

2.4.2.2. Semantic decision. In the semantic decision task AL's performance was at chance when exposure time was limited to 400 ms (French: 17/36 errors, 47%; English: 21/36 errors, 58%), with no difference between the two languages ($\chi^2 = 0.89$, ns). None of the healthy controls made any error in this task. With 1000 ms exposure, performance decreased, but remained similar for French (22/32, 69% errors) and English (24/32, 75% errors; $\chi^2 = 0.31$, ns). The number of errors was above chance due to a systematic bias of AL to classify words as belonging to the category of fruits (this bias might have been provoked by the demand of the task, which required semantic classification with only one category).

These results of the lexical decision task demonstrate that AL was partially impaired in discriminating between words and pseudowords. He was unable to determine the lexical status of plausible pseudowords, but was nearly flawless when pseudowords did not respect orthographic rules. In addition, the patient had no access to

semantic information in words even when presentation time was 1000 ms.

2.5. Language decision: words and sentences

2.5.1. Methods

Clinical observations suggested, that despite severe alexia, AL was able to determine whether a sentence or a word was written in French, English, or German. In order to examine the precision and speed of this ability, we asked him to identify the language of written words and of sentences. In the first experiment participants were asked to decide whether a word displayed for 400 ms was a French or an English word. The material employed in this experiment was composed of the same words used for the reading task.

The second experiment examined AL's language decision with sentences. Participants were asked to indicate whether a sentence displayed for 1000 ms was written in French, English, or German. We constructed 15 sentences in each language that did not contain language-specific properties, such as upper-case letters for German nouns and accents in French and German. For example, the sentence 'he is in the process of selling or renting them' was presented in English, French ('il est en train de les vendre ou de les louer') or German ('er ist dabei, sie zu verkaufen oder zu vermieten').

2.5.2. Results

AL made few errors in language decision, his performance being significantly above chance for high-frequency (8/41 errors, 19.5%; $\chi^2 = 8.4$, $p < .01$) and low-frequency words (6/46 errors, 13%; $\chi^2 = 14.6$, $p < .001$). Language decision was comparable for French words (7/36, 19% errors) and English words (7/51, 14% errors; $\chi^2 = 0.51$, ns), even when high- and low-frequency words were compared separately (high frequency: French 4/17 errors, English 3/27 errors; $\chi^2 = 1.20$; low frequency: French 3/19 errors, English 4/24 errors; $\chi^2 = 0.005$). Control participants made on average 1.7/41 errors (4.1%) for high-frequency words and 2/46 errors (4.3%) for low-frequency words. The results in sentence language identification were similar to those obtained in word language identification. Since there were three alternatives in this task, the error rate obtained by guessing would be 67%. AL made only 13.3% errors (6/45), a percentage that was significantly different from the guessing rate ($\chi^2 = 26.7$, $p < .0001$). Control participants made no errors in this task.

In order to examine whether AL's language decision was based on partial access to lexical or semantic knowledge, we compared his respective error rates in the language, lexical and semantic decision tasks. Language decision (14/87 errors, 16.1%) was far superior to lexical decision (91/204 errors, 44.6%; $\chi^2 = 21.5$, $p < .001$) and semantic decision (38/72 errors, 52.8%; $\chi^2 = 24.09$, $p < .001$), suggesting that residual lexical or semantic processing cannot account for AL's capacity to recognize written language.

AL's language decision performance strongly contrasted with his laborious reading; nevertheless, it was possible that AL relied on his impaired reading when identifying the language of words. If this were the case, we would expect to find an effect of word length on language decision (similarly to the word-length effect in reading). We compared the rates of correct answers in the language decision task for short (3–4 letters) and long (7–9 letters) words, but found no difference in French (Fisher exact test, $p = .71$) and English ($p = .55$). In order to examine whether word length affected reaction times in the language decision task as a function of language, we analyzed the data with language as categorical factor and the number of letters as covariate. Results of this analysis showed that reaction times in the language decision task were neither influenced by word length ($F = 1.68$; ns) nor by language ($F = 0.19$, ns). In addition, AL's performance in language decision was considerably faster than his performance in word reading (Fig. 2). When identifying the language of words he gave more than 90% responses within the first 4 s following target presentation, in comparison to less than 10% responses when attempting to read words. His average response time was 1148 ms for language decision against 9733 ms for reading, reflecting an increase of 748%. Control participants also found the language decision task easier, but their response time for reading compared to language decision only increased by 10%, 18%, and 66%, respectively. Their reaction time in the language decision task was neither influenced by word length ($F = 2.88$, ns) nor by language ($F = 1.38$, ns).

Considered together, the results from the reading, lexical decision, semantic decision and language decision experiments show a clear effect of task on performance. AL obtained by far the best results in language decision; he performed slightly above chance in lexical decision (though only for French words), while performance did not differ from chance in semantic categorization. The partial access of AL to information that specifies the language and partially also the lexical status of words contrasted strongly with his severely impaired reading. Considering our results in a cross-linguistic perspective, the results from the reading, lexical and semantic decision experiments suggested that performance was similar in English and French in reading, and was not affected by word frequency. Lexical and semantic decision also were similar in the two languages, with a significant effect of orthographic plausibility (plausible vs. non-plausible pseudowords), even though performance for plausible pseudowords was more severely impaired for English.

2.6. Language decision: trigrams

2.6.1. Methods

Normal readers show superior recognition of briefly presented letters when they belong to a word compared to when they are part of a nonsense letter string (McClelland & Rumelhart, 1981). This finding suggests that as a result of experience with written language, readers encode implicit rules characterizing recurring letter combinations. The fact that AL's lexical decision was strongly influenced by the orthographic regularity of pseudowords suggests that the patient's language decision might be based on the recognition of basic orthographic structure. In order to verify this hypothesis we asked the patient to identify the language of nonsense three-letter sequences (trigrams) varying in orthographic familiarity, with trigrams that had high frequency in one language and low frequency

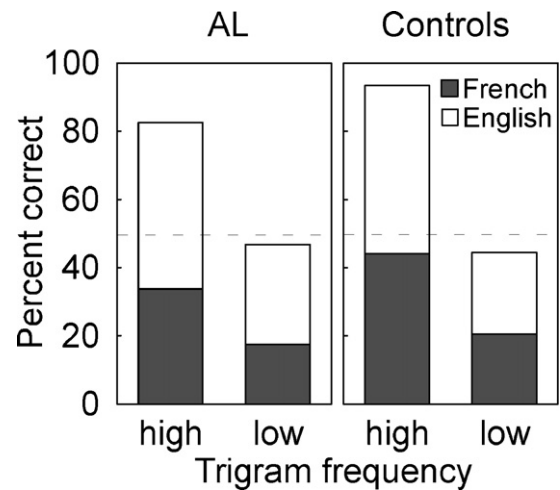


Fig. 3. Correct responses in language identification with trigrams. The dashed horizontal line indicates chance performance.

in the other language (e.g., 'iqu' in French; 'oth' in English). As a control condition, we also presented a list of trigrams with low-frequency in both languages.

In the high-orthographic familiarity condition, 20 legal high-frequency trigrams were selected in French and in English. Frequency was determined with the Lexique database (New, Pallier, Ferrand, & Matos, 2001) for French trigrams and the MCWord database (Medler & Binder, 2005) for English trigrams. French trigrams had a mean frequency of occurrence of more than 5000 per million; for English trigrams the frequency was greater than 1000 per million. In the low-orthographic familiarity condition, 15 legal low-frequency trigrams (average frequency < 500 per million in French and < 440 per million in English) were selected. None of the trigrams formed an existing word.

Trigrams were displayed for 400 ms, flanked by peripheral symbols (e.g., ##gth##) in order to give the visual impression that the trigram was embedded within a word whose first and last letters were masked. Each trigram was presented once in lowercase and once in uppercase. Language decision of high-frequency and low-frequency trigrams was tested in separate blocks. In order to increase the number of presentations, the low-frequency trigrams were tested on two different occasions.

2.6.2. Results

Fig. 3 shows the results. AL's responses were slightly biased towards classifying trigrams as English rather than French. However, independent of this bias, his language decision with high-frequency trigrams was highly accurate: he classified 66/80 (82.5%) high-frequency trigrams correctly (English: 1/40 errors, 2.5%; French: 13/40 errors, 32.5%). In contrast, performance did not significantly differ from chance for low-frequency trigrams: the patient made 64/120 (53.33%) errors for low-frequency trigrams, with a difference between English and French. The difference between high- and low-frequency trigrams was highly significant for French ($\chi^2 = 10.16$, $p < .01$) and for English ($\chi^2 = 19.13$, $p < .001$). Similarly to AL healthy participants scored well above chance for high-frequency trigrams (mean, 2.66/40 errors; 6.65%) and at chance for low-frequency trigrams (mean, 33/60 errors; 55%).

These findings provide a plausible explanation for AL's performance on language decision tasks: he showed a similar pattern of responses as healthy participants when asked to discriminate the language of high- and low-frequency trigrams, suggesting that he had relatively preserved access to basic orthographic regularities specific to each language. The question that emerges from this finding is whether AL extracted orthotactic rules by using his impaired

reading or by an alternative (faster) route. The fact that his language decision is more than seven times faster than his reading argues for the latter possibility. There is however, a further finding that argues against an explanation of AL's capacity to identify written language by using his residual reading capacity. Since AL shows a serial reading strategy, one might assume that he identifies the language of a word by systematic reading of the first characters. In analogy to our findings for high- and low-frequency trigrams this hypothesis predicts that language decision of AL is above chance for words whose initial trigram is frequent and at chance when the initial trigram has low frequency. In order to test this prediction, we extracted the initial trigram of each word used in the language decision experiment (Section 2.4) and calculated its frequency using the Lexique database (New et al., 2001) for French trigrams and the MCWord database (Medler & Binder, 2005) for English trigrams. We sorted French and English words according to the frequency of their initial trigram, split each list in half and then examined whether language decision was better for the high-frequency half than the low-frequency half. This was not the case: AL made a comparable proportion of errors when the initial trigram had high (French: 3/18 errors, 17%; English: 4/26 errors, 15%) and when it had low frequency (French: 4/18 errors, 22%; English: 3/25 errors, 12%). Thus, these findings argue against the hypothesis that AL identified the language of words based on serial reading of the initial letters.

Recent evidence suggests that the surface properties of words – termed orthographic goodness or typicality and measured by bigram or trigram frequency – influence word recognition (Hauk et al., 2006; Rogers, Ralph, Hodges, & Patterson, 2004; Woollams, Silani, Okada, Patterson, & Price, 2011). Our results suggest that AL had relatively preserved knowledge of orthographic properties as his performance in language decision on trigrams was better for high- than low-frequency trigrams. In order to perform a final analysis we examined whether a compound score reflecting orthographic typicality of whole words was a predictor of AL's performance in reading and language decision. Following a procedure described previously (Rogers et al., 2004), we extracted the frequency of each trigram that could be formed from a given word, using the corresponding French (New et al., 2001) and English databases (Medler & Binder, 2005), and then computed the sum of all trigram frequencies for each individual word.

T-tests comparing the average trigram frequency of words read correctly or classified correctly in the language decision task were not significant. Further, analysis of covariance using language as categorical predictor, and trigram frequency as continuous predictor showed that neither of these factors affected reaction time of AL (reading: $F=1.495$; ns; language decision: $F=0.32$; ns) or controls (reading $F=0.27$; ns; language decision $F=0.33$; ns) in reading and language decision. Thus, while AL was capable to determine the language of isolated trigrams based on their frequency, a compound score of within-word orthographic typicality was not a reliable predictor of reading or language classification performance.

3. Discussion

This patient with severe letter-by-letter alexia retained the ability to identify the language of a word even though he was mostly unable to identify the word through explicit reading. In contrast to his ability to detect the language of written text, he had great difficulty in reading even single letters; reading words was extremely laborious when given unlimited time and virtually impossible when exposure time was limited. To our knowledge this new form of Saffran effect – preserved access to the language of words – has previously only been reported in anecdotal form by Cohen, Dehaene, Chochon, Lehericy, and Naccache (2000).

In all experiments we assessed residual reading abilities of AL in French and English. We found that performance was highly similar in both languages: reading times and percentage of errors in reading were comparable across languages; there was no frequency effect in either language, and similar performance in the semantic decision task and the lexical decision task with non-plausible pseudowords.

An intriguing finding was that AL's language decision dissociated from his lexical decision with legal pseudowords (near chance) and from his capacity to extract semantic knowledge. Although not all LBL readers exhibit implicit reading, several patients have been described who are able to access lexical and semantic knowledge despite severely impaired reading. For example (Coslett & Saffran, 1989), reported 4 patients with pure alexia and LBL reading who performed above chance on forced-choice lexical decision and semantic categorization tasks with briefly presented words that they were unable to identify explicitly. Similarly, Lambon Ralph et al. (2004) reported a single patient who scored above chance on semantic categorization tasks despite very poor reading.

In order to account for these residual abilities (sometimes referred to as 'covert' reading) in pure alexia, Behrmann, Plaut, et al. (1998) proposed that despite impaired parallel processing of letters, some degraded information reaches higher-level (lexical/semantic) representations, which in turn provide top-down feedback and allow covert identification of written content. This hypothesis is based on the Interactive Activation Model of visual word recognition (IAM) by McClelland and Rumelhart (1981), according to which word perception takes place in a processing system with three levels of representation: the feature level, the letter level, and the word level. Activation at each level reinforces activations of the other levels through feedforward and feedback processes. Such top-down activation may explain the word-superiority effect where a letter (e.g., A) is best perceived when embedded in a word (e.g., BAG) than in a nonword (e.g., GAB). Within the IAM model, covert reading (e.g., lexical or semantic decision) is possible because visual input – albeit impoverished – reaches the word level and thus partly activates lexical and semantic representations (Behrmann, Plaut, et al., 1998). Applied to the present data this model can explain AL's impaired access to lexical and semantic representations by invoking a deficit at the lower levels (i.e., letter and possibly feature level). Indeed, AL had a severe deficit affecting performance down to the identification of single letters and which became apparent even with non-linguistic material. Therefore, his poor visual analysis might have precluded unconscious activation of higher-level representations necessary for accurate lexical and semantic judgments. In line with this account are reports of pure alexic readers whose deficit to process orthographic material extends to a general perceptual deficit (Behrmann, Nelson, et al., 1998; Farah & Wallace, 1991). Our experimental findings suggest that AL was able to extract basic orthotactic rules from words and even nonword trigrams. This finding enables us to trace a parallel to auditory word recognition. Current theories assume that the latter relies on partly independent lexical (words) and sublexical representations (phonemes and syllables; McClelland & Elman, 1986; Norris, McQueen, & Cutler, 2000). Vitevitch and Luce (1998) have suggested that the probability with which phonemes co-occur – that is probabilistic phonotactic information – facilitates classification of spoken nonwords (or unknown words) with high phonotactic probability. Orthographic structure can be operationalized as the frequency of small, meaningless letter sequences (trigrams or bigrams) in each language. High-frequency trigrams are letter-sequences whose occurrence in a given language is highly probable, while low-frequency trigrams are orthotactic patterns with low probability. Our patient was able to identify the language of frequent trigrams, suggesting that he retained the ability to detect basic orthographic regularities

that specify a given language. In lexical decision task we found that performance of AL was fast and accurate for illegal pseudowords, which was not the case for legal pseudowords.

A recent model of word identification in bilinguals (bilingual interactive activation model; Dijkstra & Van Heuven, 2002) assumes that initial stages of visual word recognition involve a parallel activation of orthographic representations in both languages. Using bilingual lexical decision tasks Lemhofer and Dijkstra (2004) have found that participants first make a distinction between the two languages and then access semantic knowledge. The authors propose that this early process of language discrimination is either based on sublexical information specific for each language (orthographic typicality) or on lexical information (orthographic neighborhood). The former hypothesis is in line with our finding that in the language decision task AL was able to categorize sublexical information specific to each language.

In conclusion, our data show that the covert ability to identify the language of written material without being able to identify words or extract lexical and semantic information depends on preserved access to sublexical orthographic representations. In normal readers the visual recognition system develops a high degree of perceptual expertise with familiar letter sequences (Gauthier, Tarr, Anderson, Skudlarski, & Gore, 1999). This knowledge concerns the linguistic regularities associated with word-parts at a sublexical level. Results of several studies have provided evidence supporting the existence of intermediate levels between letters and words in visual word processing. Using the illusory conjunction paradigm (after a presentation of a target letter, subjects are rapidly presented with a stimulus word in colored letters, and then have to indicate the color of the target letter) Rapp and Carreiras et al. (Carreiras, Vergara, & Barber, 2005; Rapp, 1992) showed that subjects were more prone to make a false color conjunction between letters which were part of the same syllable unit compared with letters that were non-syllable units. Similar results were found in ERP studies (Carreiras et al., 2005). Neuropsychological studies also found evidence that some pure alexic patients have the ability to integrate sub-lexical orthographic information. Similarly to our present study, Rosazza, Appollonio, Isella, and Shallice (2007) presented letter trigrams representing orthographically plausible or non-plausible syllables to two pure alexic patients (FC and LDS). They found that one of these patients was better able to identify the three individual letters of plausible compared to non-plausible trigrams, suggesting that access to sublexical (syllabic) information facilitated letter identification. These findings are further supported by findings from functional neuroimaging, which suggest that the perceptual system of literate adults is sensible to recurring letter combinations (Binder et al., 2006). Our patient's ability to identify written language based on trigram frequency suggests that damage to the putative occipito-temporal word-form area may severely impair reading, yet spare sublexical orthographic processing.

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