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Do patients with pure alexia suffer from a specific word form processing deficit? Evidence from 'wrods with trasnpsoed letetrs'

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ABSTRACT

It is widely accepted that letter-by-letter reading and a pronounced increase in reading time as a function of word length are the hallmark features of pure alexia. Why patients show these two phenomena with respect to underlying cognitive mechanisms is, however, much less clear. Two main hypotheses have been proposed, i.e. impaired discrimination of letters and deficient processing of word forms. While the former deficit can easily be investigated in isolation, previous findings favouring the latter seem confounded. Applying a word reading paradigm with systematically manipulated letter orders in two patients with pure alexia, we demonstrate a word form processing deficit that is not attributable to sublexical letter discrimination difficulties. Moreover, pure alexia-like fixation patterns could be induced in healthy adults by having them read sentences including words with transposed letters, so-called 'jumbled words'. This further corroborates a key role of deficient word form processing in pure alexia. With regard to basic reading research, the present study extends recent evidence for relative, rather than precise, encoding of letter position in the brain.

1. Introduction

Pure alexia is a rare neurological disorder characterised by severe reading difficulties in the absence of other language-related impairments. A pronounced increase in reading time as a function of word length (Behrmann, Plaut, & Nelson, 1998) and a compensatory letter-by-letter (LBL) reading strategy (Coslett, 2000) are the clinical hallmark features of this disorder. LBL reading is also shown by patients suffering from alexia with agraphia (e.g. Rapcsak & Beeson, 2004; Sakurai et al., 2000). However, these latter patients suffer from both reading and writing problems. Most pure alexics reported in the literature show damage to left occipito-temporal brain areas (Montant & Behrmann, 2000). Specifically, lesions affecting the paraventricular white matter of the left occipital lobe (Damasio & Damasio, 1983), the left inferior temporal and fusiform gyri (Binder & Mohr, 1992; Leff, Spitsyna, Plant, & Wise, 2006; Pflugshaupt et al., 2009), or the left occipito-temporal sulcus (Cohen et al., 2003) have been proposed as the anatomical correlate of pure alexia. The three most recent of these studies linked the common lesion site found in pure alexics with the location of the visual word form area (VWFA) in healthy adults (Cohen et al., 2003; Leff et al., 2006; Pflugshaupt et al., 2009). This suggests that the cognitive pathomechanism underlying pure alexia might have something to do with the computation or processing of the visual word form, defined as an abstract representation of ordered letter strings (Warrington & Shallice, 1980) that is invariant across changes in spatial location, case, or font (Cohen et al., 2000).

In fact, several hypotheses attributing pure alexia to impaired word form processing have been postulated. For instance, patients may have lost the ability to encode letters in parallel (Rayner & Johnson, 2005) or map the percept of all the letters in a string onto the visual word form (Leff et al., 2001). Damage to visual word forms might also cause the disorder (Warrington & Langdon, 1994). Experimental evidence in support of these hypotheses comes from tasks intended to increase the necessity for whole word reading, as opposed to sequential LBL reading. For example, patients with pure alexia showed particularly pronounced reading difficulties when words were written in script – relative to print – or when their exposure duration was very brief (Warrington & Shallice, 1980). Detecting single letters added to words, or parsing unspaced

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words also seem specifically impaired (Warrington & Langdon, 1994).

However, the idea that pure alexia results from impaired word form processing is not undisputed. Others have argued that the main deficit of patients concerns the encoding of letters on a sub-lexical level (Arguin & Bub, 1993). Indeed, many patients with pure alexia display letter confusion, which mainly affects visually similar letters (e.g. Patterson & Kay, 1982; Perri, Bartolomeo, & Silveri, 1996) and is enhanced when several letters have to be identified simultaneously (Arguin & Bub, 2005). Strong experimental support for a fundamental role of letter confusion in pure alexia comes from the observation that the characteristic increase in reading time as a function of word length – the so-called word-length effect (WLE) – can be eliminated by matching the summed confusability of constituent letters across different word lengths (Fiset, Arguin, Bub, Humphreys, & Riddoch, 2005).

In light of the latter finding, one might even ask whether difficulties in letter discrimination explain all reading problems of pure alexia patients, so that the disorder should be relabelled 'letter confusability dyslexia', as suggested by Fiset et al. (2005). Consequently, the mere existence of a specific word form processing deficit – one that is not attributable to more fundamental letter discrimination problems – is put into question. That word form processing by definition includes letter encoding considerably complicates matters. For example, it seems unclear whether pure alexics' more pronounced difficulties in reading script as opposed to print (Warrington & Shallice, 1980) should be attributed to impaired word form processing, enhanced letter confusability when words are written in a different font, or both. Investigating a specific word form processing deficit in pure alexia thus requires experimental paradigms that allow manipulating word forms while keeping letter information constant.

This requirement can be met by modifying the order of letters within words, thereby creating so-called 'jumbled words'. Provoked by a fake email message about research conducted at Cambridge University, jumbled words have recently gained attention (Grainger & Whitney, 2004; Rayner, White, Johnson, & Liversedge, 2006). The message stated that we can read them with relative ease, as long as first and last letters are in the right place. It was proposed that this 'jumbled word effect' sheds light on how the brain encodes the position of letters within words (Grainger & Whitney, 2004).

While classical models of visual word recognition assume precise-position encoding (Coltheart, Rastle, Perry, Langdon, & Ziegler, 2001; McClelland & Rumelhart, 1981) – which is at odds with the jumbled word effect and other experimental findings (e.g. Gomez, Ratcliff, & Perea, 2008) – Grainger and Whitney (2004) propose relative-position coding based on ordered letter pairs labelled 'open bigrams'. These units are open insofar as they code the position of a given letter somewhere left or right of another letter, with a maximum of two intervening letters (Grainger & Van Heuven, 2003). For instance, reading the word FOREST is assumed to activate the units representing FO, FR, FE, OR, OE, OS, RE, RS, RT, ES, ET, and ST. Analysing its jumbled variant FOERST nicely exemplifies why the open-bigram approach is considered more appropriate in explaining the jumbled word effect than precise-position encoding: the variant still activates 92% of the open bigram units (11 of 12) but only shows 67% overlap in precise letter positions (4 of 6). Jumbled word paradigms might thus be helpful not only in examining the cognitive basis of pure alexia but also in testing assumptions about how the brain encodes words.

The aim of the present study was to investigate the supposed word form processing deficit of pure alexics in three steps. First, we intended to replicate two main findings from the seminal work by Warrington and Shallice (1980), i.e. patients' enhanced difficulties when words are written in script or only briefly presented. Second, the limitation that these two difficulties seem ambiguous with regard to the underlying letter and/or word form processing deficit should be overcome by applying a jumbled word paradigm. Finally, and similar to previous studies proposing that a certain mechanism is the main cognitive deficit in pure alexia (Fiset, Gosselin, Blais, & Arguin, 2006; Rayner & Johnson, 2005), we attempted to induce LBL reading in healthy adults by having them read sentences with jumbled words while their eye movements were recorded. The task design was based on the finding that jumbling initial letters of words provokes more pronounced 'visuo-motor cost' in healthy readers than jumbling internal letters of words, relative to normal text (Rayner et al., 2006; White, Johnson, Liversedge, & Rayner, 2008).

2. Methods

2.1. Participants

Two patients (PS, RR) participated in the study. Both fulfilled the main diagnostic criteria for pure alexia: presence of a significant WLE when reading single words (Fig. 1; Table 1) in the absence of other language-related impairments. The latter was examined with an aphasia screening test, which evaluates spontaneous speech, oral repetition, reading of single words and sentences, colour and picture naming, writing of single words and sentences, auditory comprehension, and reading comprehension (Weniger, 2006). Moreover, both patients were able to identify local and global aspects of a complex visual scene as part of the same aphasia screening test (Weniger, 2006), correctly named both global and local elements of compound hierarchical letters (Navon, 1977), and were unimpaired in the identification of overlapping figures (Poppelreuter, 1917). In contrast, their reading was extremely slow – even with short function words – and characterised by overt LBL reading. Moreover, they displayed minor letter naming confusion – which exclusively concerned visually similar letters – and a trend towards prolonged reaction times during single letter naming (Table 1B). With regard to aetiology, both patients had suffered a stroke in the supply territory of the left posterior cerebral artery (PCA), causing brain damage predominantly in the ventral parts of the left occipital and temporal lobe (Table 1A). Fig. 2A depicts that the lesion of one patient (RR) included the coordinate assigned to the centre of the VWFA in healthy controls (Jobard, Crivello, & Tzourio-Mazoyer, 2003), while the other patient showed brain damage close to this coordinate.

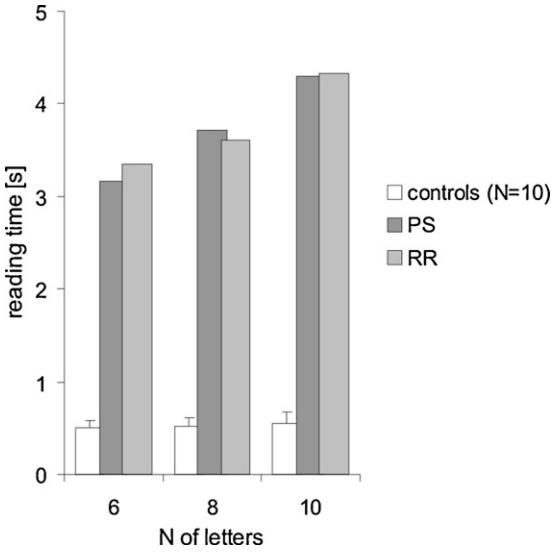

Fig. 1. Reading times from a single word reading task during which participants had to read single words of different lengths (6, 8, or 10 letters) as accurately and quickly as possible. Words were written in *Courier new* (font size: 40 pixels, corresponding to 3.72°, 4.96°, and 6.20° of visual angle) and shown one-by-one on a notebook screen placed at 60 cm from participants, to the left of a preceding central fixation point. Overall, 20 frequency-matched German nouns were displayed for every word length and presented in a pseudo-randomised sequence. Participants were instructed to press a key as soon as they identify the word, and to read the word aloud thereafter. Bars show means, error bars display standard deviations of the control group. Reading accuracy was faultless in both patients and all control participants.