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What's in a name? The characterization of pure alexia

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Pure alexia is a selective impairment of reading in the absence of other language deficits and occurs as a consequence of brain injury in previously literate individuals. The syndrome has intrigued researchers for well over a century and is the most studied of the acquired reading disorders. Pure alexia has been extensively investigated over the last 40 years within the framework of cognitive neuropsychology, but the syndrome, as a clinical entity, much predates the inferential methodology of cognitive neuropsychology. It is based on empirical generalizations by clinicians of a counterintuitive and relatively infrequent but, nonetheless, consistently observed set of behaviours in patients. However, different sets of critical features have been proposed to define the disorder. This is indicated by the range of names with similar but not identical referents with which the syndrome has been labelled over the last 120 years, e.g., *alexia without agraphia, agnosic alexia, word form dyslexia, verbal alexia, global alexia, word blindness, letter-by-letter (LBL) reading, letter-by-letter dyslexia,* and *spelling dyslexia.*

Some labels indicate degree of severity (global alexic patients are totally unable to read even single letters) while others focus on the compensatory strategies utilized by the patients (LBL-reading and spelling dyslexia). Also, concepts have differed between disciplines: In (behavioural) neurology the dissociation between impaired reading and preserved writing and language functions has been central to the definition, and thus the terms alexia without agraphia or pure alexia are commonly used. In cognitive neuropsychology, more attention has been paid to the overt or covert spelling behaviour of the patients—the word length effect in reading—and LBL-reading or LBL-dyslexia have been the terms preferred by many. Based on neuroimaging studies of patients with pure alexia (e.g., Cohen et al., 2003; Leff, Spitsyna, Plant, & Wise, 2006; Pflugshaupt et al., 2009), it has also become increasingly common to include a lesion site in ventral temporo-occipital (vOT) areas in the left hemisphere in the definition of pure alexia. Looking at published papers on pure alexia from the last few years, it is notable that most definitions include some of the features mentioned here—a word length effect, intact writing and language, and lesions affecting the left vOT—but few include all three.

A special issue of *Cognitive Neuropsychology* about pure alexia (letter-by-letter reading) was published in 1998, and in the introduction to that issue the editor, Max Coltheart, stated that "a number of important questions have emerged concerning how this disorder should be interpreted in terms of

functional models of normal reading, how studies of the disorder might contribute to the further development of such models, and what these studies might be able to tell us about the neuroanatomy of reading" (Coltheart, 1998, p. 1). He listed seven core questions and concluded his introduction saying that the works presented have "made it very clear what are the critical things we need to find out next about pure alexia" (Coltheart, 1998, p. 5). Coltheart also anticipated that methodological developments would contribute significantly to the research field, which has indeed been the case. Neuroimaging methods have contributed towards delineating the cerebral areas involved in normal and impaired word recognition (e.g., Cohen & Dehaene, 2004; Cohen et al., 2004; Leff et al., 2001; Leff et al., 2006), and computers and eyetracking devices have helped in measuring both impaired and normal reading patterns more sensitively (e.g., Behrmann, Shomstein, Black, & Barton, 2001; Pflugshaupt et al., 2009; Sheldon, Abegg, Sekunova, & Barton, 2012). The development of more rigorous statistical methods for cognitive neuropsychology have helped in more successfully dealing with a central issue in alexia research, namely the problem of individual differences in premorbid abilities (Crawford & Garthwaite, 2002; Crawford, Garthwaite, & Ryan, 2011).

There is, however, as yet no dominating consensus about how this disorder is to be explained. And, although over a 100 papers investigating pure alexia and the cerebral basis for visual word recognition have been published over the last 15 years, many of the core unresolved questions are the same as in 1998. The papers in this special issue stem from a meeting on the current status of research on pure alexia that we organized in Copenhagen, where many of these questions were discussed.

Coltheart's (1998) first and most basic question was *Is the disorder homogeneous?* by which he asks if "L-by-L reading is a syndrome in the sense of a set of symptoms which invariably co-occur and which have a single common cause" (p. 2). Coltheart uses the concept of "syndrome". But his use of the term is one of at least three current in cognitive neuropsychology. In the 1980s and 1990s, the most

popular view in the field was that a syndrome may have pragmatic clinical functions, say, for localization, prognosis, and rehabilitation purposes, but has no relevance, as a clinical generalization, for drawing theoretical conclusions about normal function (see e.g., Badecker & Caramazza, 1985; Caramazza, 1986; Caramazza & Coltheart, 2006). For such theorists and for this purpose, it is at best an index to patients whose individual performance, and that alone, is potentially of theoretical relevance for understanding normal function. A second school (e.g., Patterson & Plaut, 2009; Shallice, 1988) believe that functional syndromes can be realized in multiple patients, whose performance can reflect damage to qualitatively similar underlying systems. This perspective is a little broader than the position Coltheart adopted, as minor differences in behavioural effects can be attributed to microfunctional differences when at a more macroscopic level the lesions are functionally equivalent. While the adoption of this approach is now quite common in pure alexia research, at least three different functions have been linked to pure alexia. The oldest is the idea that it stems from damage to a visual word-form system (Cohen et al., 2000; Warrington & Shallice, 1980). Almost diametrically opposed is the idea that it stems from early prelexical damage not specific to reading (Behrmann, Plaut, & Nelson, 1998; Farah & Wallace, 1991). Somewhat intermediate between the two positions are suggestions that specific visual deficits that affect reading disproportionally are key to understanding the syndrome (Fiset, Arguin, & McCabe, 2006; Fiset, Gosselin, Blais, & Arguin, 2006; Roberts et al., 2013; Starrfelt, Habekost, & Gerlach, 2010; Starrfelt, Habekost, & Leff, 2009). Of these, the most specific is the hypothesis that the core deficit is in the ability to process "medium-to-high" spatial frequencies (Fiset et al., 2006; Roberts et al., 2013; but see, Starrfelt, Nielsen, Habekost, & Andersen, 2013), which are suggested to be important in letter and word identification, but less so for recognition of other objects. Widely different conceptions of the value of conceiving of pure alexia as a syndrome and indeed all three approaches to viewing pure alexia as a

functional syndrome are represented by papers in this special issue.

When pure alexia is treated as a functional syndrome, one behavioural feature above all others has been used as an inclusion criterion: the word length effect. This relationship between the number of letters in a word and the time taken to read is generally thought to reflect serial letter processing, or letter-by-letter (LBL) reading. One problem with using the word length effect as a defining feature is that "it could easily be the case that readers who use a compensatory letter-by-letter strategy do not all have functionally identical impairments" (Shallice, 1988, p. 80). Thus, it is not uncommon for LBL-reading to be seen in the context of surface alexia (a central alexic syndrome), for instance in patients with semantic dementia (Woollams, Hoffman, Roberts, Lambon Ralph, & Patterson, 2014). LBL-reading (or significant word length effects) may also be seen in patients with profound visual agnosia (like H.J. A.; Fiset, Arguin, Bub, Humphreys, & Riddoch, 2005; Humphreys & Riddoch, 1987), but it is quite unlikely that the functional causes of the word length effect in surface alexia and visual agnosia are the same.

It becomes very clear from the comprehensive review of the word length effect in this issue (Barton, Hanif, Björnström, & Hills, 2014) that it is not a phenomenon only observed in pure alexia. Under some circumstances, effects of word length are seen in healthy subjects (Fiset, Arguin, & Fiset, 2006); it is a characteristic of beginning reading and developmental dyslexia, and it is also a defining feature of hemianopic alexia. It has, however, commonly been taken as a defining feature of the pure alexia syndrome, and one for which theoretical interpretations have been sought. More than 20 years ago, Price and Humphreys (1992) issued a strong warning against focusing on the word length effect as a defining feature of pure alexia: "Patients who show abnormally strong word-length effects may not be reading letter-by-letter . . . , they may not have the same functional deficit and they may

not adopt the same strategy to read. Hence we suggest that it is both irrelevant and misleading to categorize them together as letter-by-letter readers" (p. 455). This warning seems to have been largely ignored by the research community (although the number of citations to the paper might seem to indicate otherwise). As an example: In 2005, in the same issue of *Psychological Science*, one paper concluded that "parallel processing contributes significantly to explicit word recognition in LBL dyslexia" (Fiset et al., 2005, p. 535), while another paper in the same issue claimed that "Letter-by-letter acquired dyslexia is due to the serial encoding of letters" (Rayner & Johnson, 2005, p. 530). While the explanations offered are clearly different, both papers define patients as letter-by-letter readers based on their word length effects and wish to explain this effect rather than the underlying deficit.

Even if it should not be used as the only diagnostic criterion for pure alexia, there seems to be general consensus that both elevated mean reaction times (RTs) and the presence of a word length effect would be central features of a functional syndrome, with more severe patients commonly spelling letters out before naming a word. Aiming to understand the relationship between generally elevated reading times and word length effects, Barton et al. (2014; BHB[1]) in the first paper in this issue, calculate a metric (effect size) that takes both variables into account. They find that although there is a systematic relationship between reading RTs and word length effects in pure alexia, the relationship is qualitatively different from that observed in healthy subjects. They report both the word length effect and the "effect size" for all published studies where data are available, providing a very important overview of previous work, which should be useful in future studies. BHB conclude in their review that "In pure alexia the word length effect . . . probably does stem from letter-by-letter reading, which may reflect not only loss of lexical whole word processing but also a variable degree of

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[1] In the following, author initials are used to refer to the papers in this special issue. For papers with more than three authors, the first three initials are used. For the single author paper, both initials of the author are used. The full reference is presented with the first use of the initials.

dysfunction in letter encoding" (Barton et al., 2014, p. 406).

Because elevated reaction times have been used as a central criterion for operationalizing a functional syndrome, most studies of pure alexia have focused on RT measures in other tasks too (e.g., Behrmann, Nelson, & Sekuler, 1998; Mycroft, Behrmann, & Kay, 2009; Roberts et al., 2013). In their contribution to this issue, Habekost, Petersen, Behrmann, and Starrfelt, 2014 (HPB) take a different approach: They investigate the ability of four pure alexic patients to report letters and words in a paradigm where accuracy in naming briefly presented stimuli, rather than response time, is the measure of interest. They compare this performance to reaction time measures for the same stimuli. In this way, they aim to investigate the visual encoding component in word and letter processing and the relationship between visual processing and naming performance. They report a somewhat complex picture of dissociations between the performance of patients with letters and words: While all patients are impaired in both visual encoding (processing speed) and naming (RT) for words, the performance with letters is comparatively less affected, particularly in the condition examining the visual processing component. This stands in contrast to the commonly held view that a deficit in (visual) single letter processing is central in explaining pure alexia. Intriguingly, the patient with normal visual letter processing speed was none the less impaired in letter naming when RT measures were employed.

In the most classical cognitive neuropsychological study in this issue, McCloskey and Schubert (2014; MS) find very similar performance between letters and numbers in their patient L.H. D. and conclude that a shared process mediates the visual identification of letters and digits. The case of number reading is particularly interesting in relation to the relative selectivity of pure alexia, as numbers, like letters, are learned symbols and are structurally very similar to letters. Since Dejerine's (1892) original observation that number reading was better preserved than letter identification in his patient Monsieur C., there

have been several reports of intact number reading in pure alexia. A recent review, however, challenged these results, by showing both that the evidence for such a dissociation is sparse, and also that normal subjects are better at perceiving digits than letters, indicating that the reported dissociation in patients may reflect a normal processing difference (Starrfelt & Behrmann, 2011). MS investigate this question in a very comprehensive case study of a patient with alexia. Their patient L.H.D. has been studied very intensively previously (McCloskey, Fischer-Baum, & Schubert, 2014; Schubert & McCloskey, 2014), and the study reported in this issue systematically approaches the question of whether the patient's deficits with digits and letters arise at the same level of processing.

The larger issue addressed is whether there are processing mechanisms that respect the categorical boundary between letters and digits, or if they are both processed by a shared system for character identification. Working within a theoretical framework specifying the levels of representation implicated in letter identification, MS convincingly show that L.H.D.'s deficits in letter and digit identification arise at the same, shared, processing level. Interestingly, they do find a dissociation between categorization of letters and digits and identification of the same stimuli: Their patient is flawless in categorizing letters and numbers as belonging to one category or the other, while being quite impaired in identifying the same stimuli. Her errors in identification do not respect the category of the stimulus, when stimuli are mixed letter and digit strings, indicating that the preserved category knowledge does not influence the identification process. MS take this as evidence for separate processes mediating character category representations and character identities and that character identification is not necessary for categorization, or vice versa. MS's study is impressive in its methodological stringency and comprehensiveness, and, as they state in their conclusion, the study goes "far beyond the demonstration of an association", but rather provides "a strong case for a single character identification process that is shared between letters and digits" (p. 458).

The question of whether pure alexia is orthography specific, or rather reflects a more general visual impairment, has long been a matter of great controversy (Behrmann et al., 1998; Gaillard et al., 2006; Mycroft et al., 2009; Roberts et al., 2013; Starrfelt et al., 2009). A recent study has also turned the problem upside down and reported preserved reading in patients with severe visual impairment, concluding that such visual deficits may not be the cause of pure alexia (Yong, Warren, Warrington, & Crutch, 2013). Woollams et al. (2014; WHR) explore this question by comparing the reading performance of a group of alexic patients with focal left occipitotemporal lesions (some of whom have pure alexia, and all of whom read letter-by-letter) to that of patients with semantic dementia, who also often show word length effects in reading (Cumming, Patterson, Verfaellie, & Graham, 2006). The two groups they investigate are matched on overall reading performance. In their analysis of reading errors in the two groups, WHR find quite different patterns of performance: a higher rate of letter-based errors in the pure alexic group and a lower rate of regularization errors. In addition, they find that although letter substitutions were common in both groups, these were characterized by visual similarity in pure alexia, while in the semantic dementia patients errors were mostly characterized by phonological similarity between stimulus and response. They interpret these findings within the framework of a connectionist model of reading, building on the primary systems view (Patterson & Lambon Ralph, 1999) and conclude that the functional loci of the reading impairment, and thus what gives rise to the word length effects in reading in pure alexia and semantic dementia, are different. They hypothesize that pure alexia is caused by a visual impairment, perhaps affecting processing of high spatial frequency information, while the reading deficit seen in semantic dementia is a result of degraded knowledge of word meaning.

While the debate about the relationship between visual processing, and visual impairments, and pure alexia has received much attention in the literature, the question of the intactness of other language functions including writing has been relatively ignored. Although the definition of pure alexia is clear—reading is impaired while writing and general language skills are unaffected—the majority of brain injuries will affect more than one function. Also, many patients displaying letter-by-letter reading have additional deficits in naming or writing, and thus patients with at least minor deficits in these domains have often been included in studies on "pure" alexia. Exploring the relation between deficits and underlying cerebral bases for reading, writing, and naming thus seems important, and two papers in this special issue do exactly that:

Purcell, Shea, and Rapp (2014; PSR) introduce a fascinating new method for lesion/symptom analysis, in their study of the interface between orthography and semantics in spelling and reading. They present three patients with deficits affecting varying aspects of reading, writing, spelling, and naming, and by analysing the intersections and dissociations between their lesions offer new insights regarding the cerebral substrate of these processes. Using the visual word form area (VWFA) as a starting point for their anatomical analyses, they specify regions posterior to this as being important in pre-lexical reading processes and also highlight an area anterior to the VWFA as being important in linking orthographic and semantic processes (OSIR—orthographic–semantic interface region). PSR hold that damage to an orthographic long-term memory system would give rise to lexical orthographic problems in both reading and spelling as it is held to store the sole representation of the orthographic form of words (it is noteworthy that pseudoword reading and spelling would be intact). A striking conclusion from their interpretation of their findings is that any clinically classic pure alexic patient whose lexical orthographic spelling is intact must have a lesion prior to any visualword form system, given that this includes the orthographic long-term memory for words. Given that many papers do not report data on spelling or writing, it is difficult to evaluate how often spelling or writing deficits accompany a lesion to the VWFA, but a quick glance at the literature suggests that at least some pure alexic patients do have

lesions not affecting the VWFA, but rather discon-
necting it from visual input (e.g., Cohen et al., 2004;
Epelbaum et al., 2008). While there are also cases
where spelling/writing is reported to be intact at
least on clinical tests in patients with lesions includ-
ing the VWFA (e.g., Starrfelt et al., 2009), it is dif-
ficult to evaluate whether more sensitive testing
might reveal lexical deficits, and this issue deserves
further attention.

While PSR's study rests on comprehensive
testing of a few patients, the study of Sebastian
et al. (2014; SGL) is notable in the number of
patients included. From a set of 234 patients
with acute left-hemisphere ischemic strokes, they
investigate the effect of lesions to Brodmann's
area (BA) 37 on reading, writing, and naming.
All patients were tested and scanned within 48
hours of their injury, and SGL use their data to
test a number of specific hypotheses regarding
the roles of medial and lateral BA 37 in lexical pro-
cessing. Their main hypothesis is that damage to
medial BA 37 (which includes the VWFA)
should affect both reading and writing/spelling,
because this is where graphemic descriptions
(invariant letter and word representations) are
computed, an idea quite similar to that presented
by PSR. Lateral BA 37, on the other hand, is
hypothesized to be involved in modality-indepen-
dent lexical access, and thus lesions here will affect
naming, regardless of the input modality. Most of
the patients with lesions to BA 37 had lesions
affecting both the medial and lateral parts, and
in these patients reading, writing, and naming per-
formance was impaired compared to controls.
Further, looking at subgroups of the patients
with lesions to BA 37, SGL find that lesions to
lateral BA 37 leave reading performance unaf-
fected (acknowledging that the tests are crude),
while naming both to visual and to tactile input
is affected. For the very few patients with impaired
reading and spelling, but intact naming, they find
that damage to medial BA 37 is important, and
indeed none of the patients with reading impair-
ment following lesions to this area showed intact
writing.

Of particular interest given the topic of this
special issue, very few of the patients showed a

pattern of performance clearly compatible with a
diagnosis of pure alexia. SGL suggest that this is
because only lesions deafferentiating medial BA
37 from visual input (but not lesions affecting this
area directly) will result in impaired reading while
leaving writing relatively spared, a conclusion
similar to that of PSR. However, as noted above,
it is not clear from the literature that writing is
necessarily affected by lesions to the VWFA/
medial BA 37, although the studies of PSR and
SGL suggest that further investigations of the
relationship between deficits in reading, writing,
and general language skills deserve further atten-
tion. It should be noted, though, that given the
acute status of the patients tested by SGL, reaction
times were not measured, so it remains unknown
whether there were patients in their sample that
read slowly but correctly, while being able to write
normally (pure alexics).

Overall, it seems that while there has been much
research concerned with pure alexia and the cogni-
tive and cerebral bases for visual word recognition
over the 15 years since the last special issue on
the topic, many of the core questions posed by
Coltheart (1998) are still unanswered. In addition
to asking whether the disorder was homogeneous,
Coltheart asked six other questions. Three are
addressed directly or indirectly in the current
volume. *"What is the locus of the patient's deficit
within a processing model of reading?"* is addressed
directly for their patient by MS. *"Is the patient's
deficit specific to reading or does it consist of a more
general visual deficit?"* is an issue that the papers
of PSR, MS, HPB, and WHR all addressed
directly or indirectly. *"What causes the patient to
read letter-by-letter?"* is central in BHB's review of
the word length effect and is also directly tackled
in Shallice's (2014; TS) paper. He argues, that at
least in most pure alexic patients, this effect arises
from a compensatory strategy. Moreover, he
makes a much more contentious argument: Given
that the word length effect arises from a compensa-
tory strategy, this has the consequence that, in
general, the behaviour of pure alexic patients
when reading words does not speak effectively to
second-generation computational models of the
orthographic word-processing stages of the word-

reading process (models that attempt to provide computational implementations of some of the processing operations involved). Thus one of Coltheart's important questions, which relates to functional models of normal reading, namely "how studies of the disorder might contribute to the further development of such models" is answered by TS for word reading in a strongly negative fashion.

For three issues discussed by Coltheart (1998), on which there was discussion in the literature before 1998, there has been relatively less work since. One of these was *what contribution the right hemisphere makes to reading (in pure alexia)*. This was initially stimulated by right-hemisphere theories of deep dyslexia (Coltheart, 1980; Saffran, Bogyo, Schwartz, & Marin, 1980) and then by theorizing on the origins of the implicit reading phenomenon in pure alexia (Coslett, Saffran, Greenbaum, & Schwartz, 1993), which relates to the second question much discussed in the 1990s of *why some pure alexia patients show implicit reading, but others do not*. The major development regarding implicit reading is a proposal by Roberts, Lambon Ralph, and Woollams (2010) that there is an inverted U-shaped relationship between severity of pure alexia and implicit reading, so that patients with reading latencies of an intermediate range should be the ones to show implicit reading effects. The question of the relative contribution of the right hemisphere is still debated, but within a different framework mostly based on (functional) imaging (Cohen et al., 2003; Henry et al., 2005; see also Barton et al., 2010). The question of lateralization may also gain further attention following a more recent conceptualization of how bilaterally distributed networks are involved in visual word recognition (Behrmann & Plaut, 2013, 2014). None of the papers in the current issue, however, centrally address these issues.

The final issue addressed by Coltheart (1998) concerns *why letter-by-letter reading . . . is affected by imageability (concreteness)*, which is the case for some pure alexia patients, at least as regards reading speed. Generally, this question has been little explored since higher order word characteristics like imageability and frequency are typically controlled in word lists presented for reading. An exception is the line of work concerned with the effect of letter confusability on LBL-reading (Fiset, Arguin, et al., 2006; Fiset, Gosselin, et al., 2006), where the complex interactions between visual and linguistic word characteristics have been explored both in alexic patients and normal subjects (see BHB for a discussion of letter confusability).

There may be many reasons for this turn in the central questions asked about pure alexia, but one reason in particular stands out from the papers in this special issue. There is little consensus on how to characterize pure alexia and even whether one should attempt to do so. Indeed a number of publications have taken the presence of a word length effect as a proxy. It should be clear from our comments above and particular those on the status of the pure alexia syndrome that we do not think questions such as "is pure alexia the result of a more general visual disorder?" and "is pure alexia caused by lesions to the visual word form area" are the same as "is letter-by-letter reading the result of a general visual disorder?" and "is letter-by-letter reading caused by lesions to the visual word form area?". In the previous special issue, these were treated as synonymous.

Although TS, as we have just pointed out, argued that analyses of the compensatory strategy (LBL-reading) cannot be expected to inform second-generation computational reading models, the more widely held view is that there is still room for studies of (pure) alexia to inform cognitive and more neuroscientifically oriented models of reading (Dehaene & Cohen, 2011; Price & Devlin, 2011). This, however, would only be meaningful if there is at least some consensus regarding how to define pure alexia and what information should be available for patients suggested to fall in this category.

We end this introduction by suggesting a definition of pure alexia, which should, in our view, be applied in future studies. We do not make this proposal for theoretical reasons but

instead to reduce the possibility of misinterpretation in the field. It is important to have a clear referent for the term _pure alexia_. We would argue that:

1. Pure alexia is an acquired reading disorder, in previously literate subjects.
2. Such patients should show correct writing (spontaneously or on dictation) and normal oral spelling.
3. They should not exhibit aphasia, dementia, or visual agnosia.
4. They should show a deficit in word reading evident in prolonged RTs and a word length effect, in reading, or be unable to read at all.[2]
5. Their lesions should be located in the posterior left hemisphere.

We advocate this rather restricted definition because of the tendency in the field to treat the existence of letter-by-letter reading as operationally equivalent to pure alexia, a tendency that the paper of BHB shows to be deeply unsatisfactory. On the other hand, we are not advocating that in order to understand pure alexia and the processes that are impaired in the condition, empirical investigations should be limited to patients that meet this list of criteria. It is clearly useful, as exemplified in this special issue, to expand the research field to include patients with associated deficits (e.g., in writing or object recognition) and to investigate them with respect to both impaired and preserved cognitive domains and the underlying cerebral substrate. Using our definition as a reference should make it clear what central aspects are impaired and preserved and hopefully aid in avoiding conceptual confusions in the future.

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[2] For an example of a "pure alexic" patient unable to read words who could still write normally see Maher, Clayton, Barrett, Schober Peterson, and Gonzalez Rothi (1998).

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