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The frequency and significance of the word length effect in neglect dyslexia

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ABSTRACT

Neglect patients often omit or misread initial letters of single words, a phenomenon termed neglect dyslexia (ND). Omissions of whole words on the contralesional side of the page during paragraph reading are generally considered as egocentric or space-based errors, whereas misreading of the left part of a word can be viewed as a type of stimulus-centred or word-based, neglect-related error. The research of the last decades shed light on several effects of word features (such as written word frequency, grammatical class or concreteness) that modulate the severity of ND. Nevertheless, almost all studies about those modulating factors were case studies and some of them have not been replicated yet. Therefore, to date we do not know how relevant such effects of different word stimuli are for a *population* of ND patients. Knowing their incidence would improve our theoretical understanding of ND and promote the development of standardized ND assessments, which are lacking so far. In particular, case studies have shown that ND error frequency increases systematically with word length (word length effect, WLE) while other single case studies found contrary results. Hence, the existence of the WLE in ND is unsettled and its incidence and significance in stroke patients is unknown. To clarify this issue we evaluated the relation between word length and the extent (number) of neglected or substituted letters within single words in ND (neglect dyslexia extent, NDE) in a group of 19 consecutive ND patients with right hemisphere lesions. We found a clear WLE in 79% (15 of 19) of our ND patients, as indicated by significant correlations between word length and NDE. Concurrent visual field defects had no effect on the WLE in our sample, thus showing no influence of early visual cortical processing stages on the WLE in neglect dyslexia. In conclusion, our results suggest a clear relationship between word length and reading errors in ND and show that the WLE is a frequent phenomenon in ND.

1. Introduction

Patients with visual neglect after unilateral right brain lesions do not report, respond, or orient to contralesional stimuli (Heilman, Watson, & Valenstein, 2012; Kerkhoff, 2001). Neglect patients typically show left-sided omissions in cancellation or visual search tasks and a rightward deviation in horizontal line bisection (Schindler & Kerkhoff, 2004; Utz, Keller, Kardinal, & Kerkhoff, 2011) as well as conspicuous reading impairments (Lee et al., 2009; Ptak, Di Pietro, & Schnider, 2012). Left-sided visuospatial neglect can impair reading in different ways. Patients typically omit entire initial words of a text line (text or space related omissions; cf. Reinhart, Keller, & Kerkhoff, 2010; Reinhart, Schindler, & Kerkhoff, 2011). Additionally, several patients omit or substitute initial letters of (horizontally presented) *single* words to the left of an identifiable "neglect point", an impairment termed neglect dyslexia (ND; Ellis, Flude, & Young, 1987; Kinsbourne & Warrington, 1962). A recent study with neglect patients and normal controls (Weinzierl, Kerkhoff, van Eimeren, Keller, & Stenneken, 2012) found that even healthy subjects frequently produced omissions and substitutions of letters, when the task difficulty was adapted by reducing presentation time of the word. In contrast to matched healthy control subjects, a high frequency of omissions and high error rates at initial (left-sided) letter positions of the word were found to be neglect-specific in this study.

Even though ND is mostly related to contralesional hemispatial neglect (further termed CN; Vallar, Burani, & Arduino, 2010) the relationship between the two disturbances is unclear. Lee et al. (2009) found the severity of CN to be a significant predictor for the frequency of ND errors. In contrast, several double dissociations have been reported in group studies (Behrmann, Black, McKeeff, & Barton, 2002) and single case studies (Cantoni & Piccirilli, 1997; Costello & Warrington, 1987; Haywood & Coltheart, 2001; Patterson

Abbreviations: ND, Neglect dyslexia; NDE, Neglect dyslexia extent; CN, Contralesional spatial neglect; WLE, Word length effect.

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& Wilson, 1990; Patterson & Wilson, 1990). In a recent investigation we found that the line bisection error (LBE) predicted the presence as well as the severity of ND, indicated by the extent of the neglected letter string and the frequency of ND errors. In contrast, we found no significant correlations between these reading-related measures and visuospatial neglect measures in a cancellation task (Reinhart, Wagner, Schulz, Keller, & Kerkhoff, 2013). Moreover, in two further investigations we found that a manipulation of the egocentric reference frame by head rotation (Reinhart et al., 2010), or optokinetic stimulation (Reinhart et al., 2011) selectively reduced *egocentric* word errors such as omissions whereas the *stimulus- or word-based* ND errors remained completely unaffected by these manipulations in the same text reading task. These results suggest two different types of reading errors: egocentric versus word-centric errors. Taken together, there is some evidence that reading impairments in neglect can occur independently in the viewer-centered (left-sided omissions of whole words) and object-centered (left-sided letter omissions/substitutions within a single word) reference frames.

To date, ND was mainly investigated in several single case reports or few group studies with small sample sizes. Therefore, the incidence of ND is unclear. In the few studies with larger sample sizes incidences of approximately 20% (Lee et al., 2009; McGlinchey-Berroth et al., 1996) up to 50% (Bisiach, Meregalli, & Berti, 1990; Ptak et al., 2012) were reported. This rather broad variance of the reported incidences might be attributable to differences in the number of presented stimuli (e.g. 12 words (McGlinchey-Berroth et al., 1996) versus 40 words (Ptak et al., 2012)). or to lexical effects of different word stimuli. The presentation of compound words where reading only the second (right) part of the compound and omitting the first (left) part of it already makes sense by itself is likely to provoke more left-sided ND errors (Ptak et al., 2012) and the use of stimuli controlled for word frequency (Lee et al., 2009) might influence error frequency in yet another way.

The research of the last decades shed light on several effects of word features that contribute to ND (for a review see Vallar et al., 2010). Nevertheless, almost all studies about those contributing factors were single cases and some of them have not been replicated yet. Therefore, to date we do not know how relevant these effects of different word stimuli are for a *population* of ND patients. Without having incidences for effects of different word stimuli, it is difficult to determine critical factors contributing to ND that could explain several dyslexic symptoms of ND patients. Knowing those factors could deepen our theoretical understanding of ND and facilitate the development of appropriate, standardized ND assessments, which are lacking so far.

1.1. Word length effect (WLE) in neglect dyslexia (ND)

Several single case studies found a correlation between word length and error frequency in ND (Behrmann, Moscovitch, Black, & Mozer, 1990; Ellis et al., 1987; Di Pellegrino, Ladavas, & Galletti, 2001; Nichelli, Venneri, Pentore, & Cubelli, 1993; Riddoch, Humphreys, Cleton, & Fery, 1990; Subbiah & Caramazza, 2000; Tegner & Levander, 1993; Vallar, Guariglia, Nico, & Tabossi, 1996). Other single case studies reported contrasting results (Behrmann et al., 1990; Cantoni & Piccirilli, 1997; Costello & Warrington, 1987; Hillis & Caramazza, 1991; Miceli & Capasso, 2001). To the best of our knowledge, only one group study evaluated the word length effect on ND error frequency in seven brain damaged patients. Takeda & Sugishita, (1995) reported that only two of their seven patients had a (non significant) tendency to make more ND errors in longer words. However, as the authors noted, only these two patients made enough errors at all in the 50 presented words in order to enable a statistical analysis of this issue.

In most studies ND error frequency was exclusively used as a measure of the severity of ND investigating the word length effect. A word length effect on the *position of the neglect point* within the word as another measure of ND severity has been reported merely in four case studies (Ellis et al., 1987; Hillis & Caramazza, 1991; Riddoch et al., 1990; Tegner & Levander, 1993). To our knowledge, the only study investigating the relation between the position of the neglect point and word length in a large sample size ($n=64$) ND was that of Schwartz, Ojemann, and Dodrill (1997) in patients with complex partial seizures receiving a right hemisphere injection of sodium amobarbital. The authors reported a significant correlation of $r=0.65$ between the position of the neglect point in a word and the length of the letter string in this sample. However, no study has investigated this issue in a larger sample of *stroke patients* with ND.

All in all, the significance and incidence of the WLE in ND in right hemisphere stroke is unclear at present. Therefore, in this study we investigated the effect of word length on the *spatial extent of the neglected initial letter string* (termed neglect dyslexia extent; NDE), in a larger group of 19 consecutive ND patients with right hemispheric stroke. The obtained results would inform us about whether the word length is a critical factor for increasing the probability of ND errors.

2. Methods

2.1. Subjects

19 consecutive patients (seven female; mean age=60.79 years; SD=9.41) all with a single right-hemispheric, vascular brain lesion (mean time post lesion=14.21 weeks; SD=7.50) and moderate (at least 1.03% ND errors) to severe left-sided ND were included (Table 1). All patients had a moderate to severe left-sided contralesional spatial neglect according to the results of two tests (star cancellation and bisection of a 20 cm long horizontal line) comparable to those of the behavioral inattention test (Halligan, Marshall, & Wade, 1989; Wilson, Cockburn, & Halligan, 1987). All patients had a visual acuity of at least 0.80 (=80%) for the near viewing distance (0.4 m) – in which all experimental reading tasks were performed – and at least 9 years of education. The study was approved by the ethics committee of the Ludwigs-Maximilian-Universität, München/Germany, Project Nr. 352-09 in November 2009. The experiment was conducted in accordance to the Declaration of Helsinki II. All patients gave their informed consent prior to investigation. Binocular visual fields were mapped perimetrically with a Tübingen or Goldmann perimeter in all patients (details see Kuhn, Heywood, & Kerkhoff, 2010; results see Table 1). Dynamic visual perimetry was performed with a circular white target (Tübingen perimeter, luminance: 102 cd/m^2; size: 1.02°) or the V4 stimulus (Goldmann perimeter) in a completely dark room (see Weinzierl et al., 2012 for a more detailed description).

2.2. Experimental reading tests

We employed 45 short reading texts of different lengths (mean length: 51.7 words, range: 43–65; arranged in 8–10 lines) containing words of 2–11 letters from two story books (see Reinhart et al., 2010, 2011). The margins of each text were irregularly indented on both sides (see example in Fig. 1A). Indented paragraph reading tasks are a highly sensitive measure of the reading performance in neglect patients (Bachman, Fein, Davenport, & Price, 1993; Caplan, 1987; Towle & Lincoln, 1991) and are not confounded by differences in years of schooling (Bachman et al., 1993). Eight to ten words on every margin (left and right side) of each text were filler words (for example words as “a”, “but”, “in”, very”, often”, etc.) and were not necessary for understanding the semantic context of the text. The main message of the text could be understood even if most or all of these filler words were omitted. All texts were matched with respect to length (number of words, letters and lines) and spatial arrangement. The number of words displayed on each side of the reading texts was balanced (mean length left: 25.8 words, mean length right: 26.00 words). There was no statistical difference between the number of words presented on the left and right text side when all 45 texts were compared [$t(44)=0.34$, $p=0.76$]. The texts were displayed sequentially one by one within an $8\times24°$ large rectangular white field on a 17″—computer screen in 0.4 m distance to the observer. Texts were presented in black print (Arial, capital letters, point size 22) on a white background at a distance of 0.4 m to the patient's eyes. The midline of the computer screen was perpendicular to the patients' trunk midline.

Subjects were instructed to read out aloud five to eight of the 45 texts displayed sequentially on the screen. These texts were presented within one session of