



Research paper

A longitudinal linguistic analysis of written text production in a case of semantic variant primary progressive aphasia



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ABSTRACT

The semantic variant of primary progressive aphasia (svPPA) presents with a degradation of semantic knowledge due to atrophy of the anterior temporal lobe and is characterized by impaired confrontation naming and impaired single-word comprehension. So far, little is known about the development of symptoms and their order of occurrence in the pre-clinical phase, and information regarding written text production is scarce.

We had the unique opportunity to analyze the diary of a man written over a time span of 12 years before he was diagnosed with svPPA. We sought to identify the earliest indicators of cognitive change in his diary entries, and to track the important changes over time.

Based on transcripts of the entries (one week every six months) we assessed the overall structure, vocabulary, surface dysgraphia and semantic paraphasia, syntax, and morphology. We found changes in all domains up to seven years before the clinical diagnosis. The earliest changes concerned the vocabulary, with decreased variety and increased use of high frequency words. This was followed by syntactic and morphological errors. We found no increase of surface dysgraphia. Semantic paraphasias increased only during the last three years but characterized the entries of the last year.

We were therefore able to further corroborate recent findings regarding difficulties in the morpho-syntactic domain in this patient group. In this natural context for written text production, such errors seem, in addition to changes in vocabulary, to be the first error types to appear, possibly as a result of compensating for the degradation of semantic representations.

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1. Introduction

Dementia, a decline of mental abilities that interferes with activities of daily life, is caused by various neurodegenerative pathologies and can feature language impairments. Whereas Alzheimer's dementia (AD), the most common form of dementia, is characterized by memory impairments, and language impairments usually emerge only during disease progression, the latter are the core symptoms in a group of syndromes termed primary progressive aphasia (PPA) first proposed by Mesulam (1982).

The current consensus (Gorno-Tempini et al., 2011) differentiates three forms of PPA: 1) the non-fluent/agrammatic variant (nfvPPA), 2) the logopenic variant (lvPPA) and, 3) the semantic variant (svPPA, also known as semantic dementia). The prominent abnormalities in svPPA concern semantic memory function and result from atrophy of the anterior temporal lobes that causes a degradation of semantic representations (Jefferies & Lambon Ralph, 2006; but see Bright, Moss, Stamatakis, & Tyler, 2008). This degradation leads to impaired confrontation naming and impaired single-word comprehension, which are core and obligatory symptoms of svPPA (Gorno-Tempini et al., 2011). In naming, patients may make coordinate or superordinate errors, which is qualitatively different to individuals with stroke aphasia (Jefferies & Lambon Ralph, 2006). Other indicators of svPPA are surface dyslexia or dysgraphia (i.e., regularization of orthographically irregular words), spared repetition, and spared speech production.

However, as pointed out by Sajjadi, Patterson, Tomek, and Nestor (2012), the diagnostic examination is mostly conducted at the single word level and inaccurate conclusions might be drawn about a patient's performance at the discourse/text level. The spoken language performance of svPPA patients is described as fluent but empty, full of overlearned phrases and semantic paraphasias (i.e., replacement of the target by a semantically related item) (Reilly, Rodriguez, Lamy, & Neils-Strunjas, 2010; Warren, Rohrer, & Rossor, 2013). To a certain extent, connected speech can sound quite normal (Sajjadi et al., 2012) but patients tend to lose their differentiated vocabulary over time and use more general and more frequent instead of semantically precise words (Ash et al., 2006; Bird, Lambon Ralph, Patterson, & Hodges, 2000; Bozeat et al., 2003; Fraser et al., 2014; Laisney et al., 2011). Apart from the changes in vocabulary, errors of syntax and morphology may also occur in the spoken language of individuals with svPPA, as has – somewhat contrary to initial expectations – been pointed out recently (Auclair-Ouellet, 2015; Benedet, Patterson, Gomez-Pastor, & la Rocha, 2006; Gorno-Tempini et al., 2011; Kavé, Leonard, Cupit, & Rochon, 2007; Leyton & Hodges, 2014; Meteyard & Patterson, 2009; Meteyard, Quain, & Patterson, 2014; Silveri et al., 2014; Thompson et al., 2012; Wilson et al., 2010, 2014). Individuals with svPPA seem to show syntactic abnormalities in the sense of paragrammatic constructions such as sentence inversions or duplications and substitutions on the morphological level. Error rates are usually higher for atypical, less familiar or infrequent constructions.

Despite the wide-ranging investigation of language impairments associated with dementia that started more than 30 years ago (Appell, Kertesz, & Fisman, 1982; Mesulam, 1982; Snowden, Goulding, & Neary, 1989; Warrington, 1975) two aspects are still relatively neglected. First, there is a lack of knowledge about the development of symptoms and their order of occurrence in the preclinical phase of different types of dementia, and this is especially true of the less well-known syndromes. Second, written text production has barely been explored in patients with dementia.

Only three studies have, to the best of our knowledge, investigated the written text production of individuals later diagnosed with AD. Two studies conducted computer-based analyses of prose texts by professional writers. The analysis of renowned Irish author Iris Murdoch's work (Garrard, Maloney, Hodges, & Patterson, 2005) revealed an increased use of higher frequency words, a less varied vocabulary, and a shallower and more banal plot in her last novel, which was written around the time AD was diagnosed, four years before her death. Word length, use of different word classes, the overall structure of the texts, and syntactic composition did not differ between three novels from the beginning, prime and end of her writing career. Similar results and additional evidence for difficulties in finishing the novel (writer's block) were found in the analysis of texts from the Dutch writer Gerard Reve, who also suffered from AD (van Velzen & Garrard, 2008). Abnormalities of language use in written text production were detectable years before the clinical diagnosis of dementia. This was also the case in a study by Snowdon et al. (1996), the so-called nun study. The analyses of a single, hand written page produced about 58 years earlier, enabled the researchers to identify the nuns later affected by AD. Their texts showed a lower rate of idea density (i.e. the quantity of propositions/ideas per sentence) and a lower degree of syntactic complexity. The nuns were not professional writers, which might explain why lower syntactic complexity was found very early in their texts but not in Murdoch's and Reve's work.

To date there do not appear to have been any longitudinal investigations of written text in svPPA. In the present study, we had the opportunity to analyze the written texts of a man, HK, who was diagnosed with svPPA 12 years after he began to write daily diary entries. We studied the texts to identify the onset of linguistic abnormalities, and to document the "linguistic fingerprint" of the disease in this individual over time. We also considered whether HK's written language would parallel the patterns found in the spoken language production of individuals with svPPA, and to what extent the changes in his written texts would be comparable to changes reported in the written language of individuals with probable AD. Our analyses included the overall structure, vocabulary, surface dysgraphia and semantic paraphasia, syntax, and morphology.

On the basis of the findings regarding spoken language production in individuals with svPPA and written language production by individuals with AD cited above, we expected various changes on different levels. Regarding the overall structure, we expected no change in the composition of the diary entries but a decrease in the number and length of entries, and a trivialization of their topics. Furthermore, we expected that HK's diary entries would show a decreasing variety of vocabulary over time as well as an increased use of higher frequency words. As surface dysgraphia and semantic paraphasia

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