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Frontal dynamic aphasia in progressive supranuclear palsy: Distinguishing between generation and fluent sequencing of novel thoughts

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ABSTRACT

Frontal dynamic aphasia is characterised by a profound reduction in spontaneous speech despite wellpreserved naming, repetition and comprehension. Since Luria (1966, 1970) designated this term, two main forms of dynamic aphasia have been identified: one, a language-specific selection deficit at the level of word/sentence generation, associated with left inferior frontal lesions; and two, a domaingeneral impairment in generating multiple responses or connected speech, associated with more extensive bilateral frontal and/or frontostriatal damage. Both forms of dynamic aphasia have been interpreted as arising due to disturbances in early prelinguistic conceptual preparation mechanisms that are critical for language production. We investigate language-specific and domain-general accounts of dynamic aphasia and address two issues: one, whether deficits in multiple conceptual preparation mechanisms can co-occur; and two, the contribution of broader cognitive processes such as energization, the ability to initiate and sustain response generation over time, to language generation failure. Thus, we report patient WAL who presented with frontal dynamic aphasia in the context of progressive supranuclear palsy (PSP). WAL was given a series of experimental tests that showed that his dynamic aphasia was not underpinned by a language-specific deficit in selection or in microplanning. By contrast, WAL presented with a domain-general deficit in fluent sequencing of novel thoughts. The latter replicated the pattern documented in a previous PSP patient (Robinson, et al., 2006); however, unique to WAL, generating novel thoughts was impaired but there was no evidence of a sequencing deficit because perseveration was absent. Thus, WAL is the first unequivocal case to show a distinction between novel thought generation and subsequent fluent sequencing. Moreover, WAL's generation deficit encompassed verbal and non-verbal responses, showing a similar (but more profoundly reduced) pattern of performance to frontal patients with an energization deficit. In addition to impaired generation of novel thoughts, WAL presented with a concurrent strategy generation deficit, both falling within the second form of dynamic aphasia comprised of domain-general conceptual preparation mechanisms. Thus, within this second form of dynamic aphasia, concurrent deficits can co-occur. Overall, WAL presented with the second form of dynamic aphasia and was impaired in the generation of novel thoughts and internally-generated strategies, in the context of PSP and bilateral frontostriatal damage.

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1. Introduction

Frontal dynamic aphasia is characterised by a severe impairment in propositional language that results in profoundly reduced, but relatively normal, speech output. Patients with dynamic

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aphasia rarely initiate conversation and have difficulty conveying a narrative or story particularly when elaboration or formulation of thought is required (Luria, 1966, 1970). This severe reduction of spontaneous speech occurs despite well-preserved core language skills (naming, repetition, comprehension, and reading), with articulation and syntax either intact or poor (*pure* and *mixed* dynamic aphasia, respectively; for review see Robinson et al. (2005, 2006). Luria coined the term *frontal dynamic aphasia*, to distinguish one form of transcortical motor aphasia that affected the impulse to speak rather than reflecting a disturbance to core







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language skills, which typically gives rise to the term 'aphasia'. Similar to many since Luria, we adopt the term *frontal dynamic aphasia* as he was the first to provide a theoretical account of the underlying mechanism; namely an inability to translate a plan into a linear scheme of a sentence (Luria, 1970, 1973; Luria and Tsvet-kova, 1967). The intention or plan existed; however, reduced propositional speech resulted from a failure to form a linear scheme.

Investigations of propositional speech failures since Luria have resulted in the identification of two main forms of dynamic aphasia, one language-specific and one domain-general (detailed in Robinson et al. (2006)). Broadly, investigations of both the language-specific and domain-general forms of dynamic aphasia allow the specification of the underlying mechanisms crucial for generating propositional language. These mechanisms operate at an early stage of speech production, prior to lexical selection and grammatical encoding, and are focussed on prelinguistic conceptualisation of a message, termed the conceptual preparation stage (Levelt, 1989, 1999; Sherratt, 2007). In Levelt's model the initial conceptualiser stage involves the generation of a new conceptual structure or message to be subsequently realised as overt speech. At this stage, a speaker attends to the current topic or focus, shifts their attention to new topics as the communicative context demands, and monitors conversation. Along similar lines, Sherratt (2007) proposed that at this prelinguistic stage the conceptual framework for a message is generated through a process of inserting and integrating semantic information, selecting and prioritising information for expression, and selecting and sequencing the necessary thoughts to reflect the message to be expressed. In both the Levelt and Sherratt models, conceptual preparation results in a preverbal message that is not yet linguistic but contains the necessary conceptual structure required for linguistic formulation and articulation. Disruption to these early conceptual preparation mechanisms has been thought to give rise to both the language-specific and domain-general forms of dynamic aphasia.

1.1. Language-specific accounts: the first form of dynamic aphasia

The first form of dynamic aphasia is characterised by languagespecific deficits at the level of word and sentence generation, and is associated with left inferior frontal damage. In addition to Luria's account of dynamic aphasia, there have been two main language-based accounts; namely, an inability to select from amongst competing verbal responses (Robinson et al., 1998, 2005) and impaired verbal planning that has similarities to Luria's account (Costello and Warrington, 1989). My colleagues and I specifically investigated the verbal planning account in several dynamic aphasia cases using Costello and Warrington's critical sentence construction test, which requires single words to be arranged into a meaningful and grammatical sentence (e.g., pond/the/frozen/was \rightarrow The pond was frozen). Our investigations did not support a verbal planning explanation and, instead, resulted in the proposal of a language-specific selection account. When patients were given a series of experimental tests, they failed to generate a single word or sentence only when a stimulus activated many, compared with a dominant or few, response options that compete for selection (Robinson et al., 1998, 2005; see also Crescentini, et al. (2008) and Robinson (2013)). This selection deficit is at a conceptual response (or proposition) level, as distinct from a stimulus-response association level in the Stroop Test (further discussed in Robinson (2013)). An inability to select from amongst competing conceptual responses has been reported in several patients with left inferior frontal damage who passed the Costello and Warrington verbal planning test (OTM-Crescentini et al., 2008; ANG & CH-Robinson et al., 1998[,] 2005; MC-Robinson, 2013). However, we note that

after most of these cases were reported, Bormann et al. (2008) investigated 'verbal planning' with several newly devised microplanning tests, although they interpreted their patient's dynamic aphasia as being due to macroplanning rather than microplanning difficulties. Macroplanning and microplanning are distinct core prelinguistic conceptual preparation processes (Levelt, 1989, 1999). Macroplanning is the process by which the speaker will focus attention on a specific message (current focus), choose what to say next (discourse focus) and in what order (sequencing), which is also impacted by broader process-related factors such as working memory or attention limitations. A very simple example is detailed by Levelt (1999, p. 90) that involves a speaker being shown an array of shapes in spatial relation to each with the goal of informing another about the layout of the pattern. Of note, this has similarities to the complex scene description tasks frequently used to elicit propositional speech. By contrast, microplanning is language-dependent and requires the speaker to determine the informational perspective of the conceptual information (e.g., the following statements contain the same conceptual information but are from different perspectives, 'there is a house with a tree to the left of it' or 'there is a tree with a house to the right of it' Levelt (1999, p. 91)). In sum, verbal (or *micro*) planning may have a role in propositional speech generation, in addition to selection, but the original Costello and Warrington (1989) critical sentence construction test may have lacked sensitivity for detecting subtle verbal planning impairments. Thus, in the current study we will investigate both the selection and microplanning language-specific mechanisms that operate at the level of sentence generation and characterise the first form of dynamic aphasia.

1.2. Domain-general accounts: the second form of dynamic aphasia

The second form of dynamic aphasia is not as well characterised but has been documented in patients with domaingeneral deficits and bilateral frontal and/or subcortical damage. These patients typically perform well on single word/sentence generation tests but impairments become apparent when required to generate multiple items as is the case for connected speech (i.e., discourse) rather than a sentence, or for fluency tasks that involve a single cue (verbal or non-verbal). Domain-general accounts of dynamic aphasia have included deficient semantic strategy forma*tion/use* to search the lexical-semantic network (Gold et al., 1997) and a failure to spontaneously activate or initiate lexical-semantic representations (Cox and Heilman, 2011; Raymer et al., 2002; for similar account see Satoer et al. (2014)), which have also been shown for verbal and non-verbal responses. However, a case with dynamic aphasia and a language-specific selection deficit is on record with intact strategy formation/use and non-verbal generation (CH-Robinson et al., 2005). A further domain-general account of dynamic aphasia was proposed on the basis of a patient (KAS) with progressive supranuclear palsy (PSP). KAS's performance on selection and verbal planning tasks was intact but she was impaired on discourse generation and non-verbal generation tasks. KAS's reduced propositional speech was attributed to a deficit in the fluent sequencing of novel thoughts (Robinson et al., 2006; for similar account see Bormann et al. (2008)). Recently, this mechanism was suggested to comprise two distinct processes: idea (or novel thought) generation and fluent sequencing of ideas (Robinson, 2013). An idea generation deficit manifests as a reduction in quantity whereas a *fluent sequencing* deficit manifests in perseveration or repetition of ideas. In some respects the idea generation component resembles a failure to spontaneously activate or initiate verbal and non-verbal responses as both result in a paucity of ideas (e.g. Raymer et al., 2002; Robinson, 2013). The damage associated with domain-general accounts has typically been widespread, encompassing bilateral frontal and/or subDownload English Version:

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