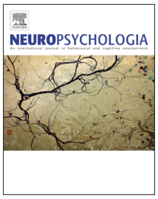
Neuropsychologia 86 (2016) 93–102

Contents lists available at ScienceDirect

Neuropsychologia

journal homepage: www.elsevier.com/locate/neuropsychologia



When will a stuttering moment occur? The determining role of speech motor preparation

Sarah Vanhoutte [a,b,*], Marjan Cosyns [b], Pieter van Mierlo [c], Katja Batens [d,e], Paul Corthals [b,f], Miet De Letter [b,d], John Van Borsel [b], Patrick Santens [a,d]

[a] *Department of Internal Medicine, Neurology, Ghent University, Ghent University Hospital, De Pintelaan 185, 1K12A, B-9000 Ghent, Belgium*
[b] *Department of Speech, Language and Hearing Sciences, Ghent University, Ghent University Hospital, De Pintelaan 185, 2P1, B-9000, Ghent, Belgium*
[c] *MEDISIP, Department of Electronics and Information Systems, Ghent University – iMINDS Medical IT Department, Ghent University Hospital 185, 5-Blok B, B-9000 Ghent, Belgium*
[d] *Department of Neurology, Ghent University Hospital, De Pintelaan 185, 1K12A, B-Ghent, Belgium*
[e] *Department of Physical Therapy and Motor Rehabilitation, Ghent University Hospital, De Pintelaan 185, B-Ghent, Belgium*
[f] *Faculty of Education, Health and Social Work, University College Ghent, Keramiekstraat 80, B-9000 Ghent, Belgium*

ARTICLE INFO

Article history:
Received 28 July 2015
Received in revised form
28 February 2016
Accepted 18 April 2016
Available online 19 April 2016

Keywords:
Contingent negative variation
Stuttering severity
Stuttering frequency
Compensation
Motor preparation
Basal ganglia

ABSTRACT

The present study aimed to evaluate whether increased activity related to speech motor preparation preceding fluently produced words reflects a successful compensation strategy in stuttering. For this purpose, a contingent negative variation (CNV) was evoked during a picture naming task and measured by use of electro-encephalography. A CNV is a slow, negative event-related potential known to reflect motor preparation generated by the basal ganglia-thalamo-cortical (BGTC) – loop. In a previous analysis, the CNV of 25 adults with developmental stuttering (AWS) was significantly increased, especially over the right hemisphere, compared to the CNV of 35 fluent speakers (FS) when both groups were speaking fluently (Vanhoutte et al., (2015) doi: 10.1016/j.neuropsychologia.2015.05.013). To elucidate whether this increase is a compensation strategy enabling fluent speech in AWS, the present analysis evaluated the CNV of 7 AWS who stuttered during this picture naming task. The CNV preceding AWS stuttered words was statistically compared to the CNV preceding AWS fluent words and FS fluent words.

Though no difference emerged between the CNV of the AWS stuttered words and the FS fluent words, a significant reduction was observed when comparing the CNV preceding AWS stuttered words to the CNV preceding AWS fluent words. The latter seems to confirm the compensation hypothesis: the increased CNV prior to AWS fluent words is a successful compensation strategy, especially when it occurs over the right hemisphere. The words are produced fluently because of an enlarged activity during speech motor preparation. The left CNV preceding AWS stuttered words correlated negatively with stuttering frequency and severity suggestive for a link between the left BGTC – network and the stuttering pathology. Overall, speech motor preparatory activity generated by the BGTC – loop seems to have a determining role in stuttering. An important divergence between left and right hemisphere is hypothesized.

1. Introduction

Stuttering is a speech disorder in which the smooth succession of speech sounds is interrupted by the repeated occurrence of prolongations, blocks and repetitions of sounds and/or syllables. When stuttering is of developmental origin, manifesting itself for the first time during childhood, it is called developmental stuttering (Bloodstein and Ratner, 2008). About 95% of children who stutter start to stutter by the age of 4 years (Yairi and Ambrose, 2005). One of the neurological characteristics of developmental stuttering is abnormal speech motor preparation. Speech motor preparation contains all processing stages in which a phonological word is transferred into concrete, context-specific articulatory motor commands (Peters et al., 2000; Indefrey and Levelt, 2004;

---

[*] Corresponding author at: Department of Speech, Language and Hearing Sciences, Ghent University, Ghent University Hospital, De Pintelaan 185, 2P1, B-9000, Ghent, Belgium.

E-mail addresses: sarah.vanhoutte@ugent.be (S. Vanhoutte), marjan.cosyns@ugent.be (M. Cosyns), pieter.vanmierlo@ugent.be (P.o.s. van Mierlo), katja.batens@ugent.be (K. Batens), paul.corthals@ugent.be (P. Corthals), miet.deletter@ugent.be (M. De Letter), john.vanborsel@ugent.be (J. Van Borsel), patrick.santens@ugent.be (P. Santens).

http://dx.doi.org/10.1016/j.neuropsychologia.2016.04.018
0028-3932/© 2016 Elsevier Ltd. All rights reserved.

Indefrey, 2011). Its major cortical neuroanatomical substrates are the ventral premotor cortex (vPMC) and the adjacent and partly overlapping inferior frontal gyrus (IFG) which includes in the left hemisphere, the well-known Broca's area (Brodmann area 44, 45). Subcortically, the thalamus and basal ganglia are of vital importance as well. They will form a reciprocal loop with the vPMC called the basal ganglia-thalamo-cortical (BGTC) – loop.

Several theories propose a key role for speech motor preparation in stuttering (Alm, 2004; Howell, 2004; Giraud et al., 2008; Civier et al., 2013). In addition, many neuroimaging studies report positive correlations between stuttering measures and neural activity in several structures of the BGTC – loop (Braun et al., 1997; Fox et al., 2000; Giraud et al., 2008; Chang et al., 2009; Kell et al., 2009; Ingham et al., 2012). Neurophysiological studies evidence the importance of motor preparation as well. They highlight dysfunctions in the transmission of sensorimotor programs to the motor cortex, particularly in the left hemisphere (Salmelin et al., 2000; Neef et al., 2015b).

Motor preparation can also be evaluated with electro-encephalography (EEG) by evoking an event-related potential (ERP) called the Contingent Negative Variation (CNV). The CNV is a slow, negative ERP occurring in between two successive stimuli. The first stimulus (S1) is a warning stimulus which precedes the second, called imperative, stimulus (S2). This S2 requires a motor response (Walter et al., 1964; Rohrbaugh and Gaillard, 1983; McCallum, 1988; Regan, 1989; Golob et al., 2005). When the interstimulus interval is ≥ 2 s, an early and a late CNV can be distinguished. The early CNV occurs within the first second following S1 and is related to orientation. It is the late CNV, occurring just before S2, that primarily represents motor preparation (Walter et al., 1964; Loveless and Sanford, 1974; Rohrbaugh and Gaillard, 1983; McCallum, 1988; Regan, 1989). Its amplitude/slope is generally accepted to reflect the amount of neural activity within the BGTC – loop (Lamarche et al., 1995; Hamano et al., 1997; Gomez et al., 2003; Bares et al., 2007; Fan et al., 2007).

In a recent EEG study from our laboratory, speech motor preparation was evaluated in adults with developmental stuttering (AWS) by use of a CNV evoking picture naming task (Vanhoutte et al., 2015). The late CNV was found to be significantly increased for the AWS compared to the fluent speakers (FS) suggesting a significant increase in BGTC – loop activity prior to speech production. This increase correlated positively with stuttering frequency and severity. Remarkably, the increased activity during speech motor preparation occurred preceding fluently produced single words. Two explanations may account for the fluent word production: (1) isolated word production is known to evoke no or only a little stuttering (Brown, 1938; Adams et al., 1973) probably due to its low demands on the neural speech motor system (Bloodstein and Ratner, 2008). Thus, when only a limited load is imposed on the speech motor system, motor preparation dysfunctions are either not enough to evoke stuttering or can be overcome by another system. (2) The words were produced fluently *because* an enlarged activation during speech motor preparation was present.

These two opposite interpretations are related to a longstanding discussion concerning the cause and the consequence of lifelong stuttering. Because developmental stuttering starts during childhood, neuroanatomical growth and maturation of children who stutter may follow an abnormal trajectory (Chang, 2011; Beal et al., 2013, 2015). Moreover, the brain will try to overcome these deficiencies by causing neural adaptation and compensatory processes that will further shape structural development (Chang et al., 2015). As a result, the neural activity and morphology pattern observed in AWS is a combination of the cause of stuttering on the one hand and compensation strategies and the consequence of stuttering on the other hand.

It is an ongoing discussion which neural anomalies are related to the cause and which to consequence/compensation. Particularly the relative role of left and right hemisphere has been addressed. A recent meta-analysis made a distinction between neural findings related to fluent and related to stuttered speech in AWS (Belyk et al., 2014). An increased activation of the right frontal operculum (RFO) was only observed during fluent speech supporting the suggestion by Preibisch et al., (2003) that RFO overactivation is a successful compensation strategy enabling fluent speech. Anomalous increased brain activity in one hemisphere may reflect a compensation for disturbed signal transmission in the contralateral hemisphere. Indeed, right IFG is involved in inhibiting speech acts that are generated by the left IFG (Xue et al., 2008) and would only interfere when left IFG experiences problems (Lu et al., 2010). A decreased fractional anisotropy (FA) of the white matter underneath left ventral sensorimotor cortex, closely located to left IFG, has repeatedly been reported (Sommer et al., 2002; Chang et al., 2008; Watkins et al., 2008; Cykowski et al., 2010; Connally et al., 2014). Moreover, a recent meta-analysis on diffusion tensor imaging data observed an additional cluster of decreased FA along the superior longitudinal fasciculus, also in the left hemisphere (Neef et al., 2015a). These findings support the hypothesis that left hemisphere abnormalities may be associated with the neural basis of stuttering and that right hemisphere deficits are related to adaptation and compensation strategies.

The increased CNV found in our study showed a laterality aspect as well (Vanhoutte et al., 2015). Although motor preparation was bilaterally increased with respect to stimulus onset (i.e. stimulus-locked or S-locked analysis), it was only significantly increased over the right hemisphere with respect to lip movement onset as measured by electromyography (EMG) of the orbicularis oris muscle (i.e. response-locked or R-locked analysis). As the R-locked analysis takes reaction time into account, activities related to response execution would be more pronounced in the R- than in the S-locked analysis (Riès et al., 2013). The results of the R-locked analysis are thus slightly in favour of the compensation hypothesis.

To clarify this “cause-compensation” discussion for the CNV results, it may be interesting to evaluate speech motor preparation preceding stuttered words. If the increased motor preparation prior to fluently produced words is related to successful compensation, speech motor preparation prior to stuttered words would be significantly lower than speech motor preparation prior to fluent words. Neurological research on stuttered speech is extremely scarce because AWS mainly speak fluent in experimental settings. Recently, two meta-analyses compared the neural correlates of stuttered speech with natural fluent speech (Belyk et al., 2014) and natural and induced (e.g. choral speech) fluent speech (Budde et al., 2014) in AWS. Both meta-analyses associated stuttered speech with an increased activation in the cerebellum and the supplementary motor area and a decreased activation in the superior temporal gyrus. Unfortunately, the majority of the included studies referred to stuttered speech that is embedded in fluent speech with percentages of stuttered syllables (% SS) starting from as low as 2.5%.

To our knowledge, only four studies compared 100% stuttered with 100% natural fluent speech in AWS (Jiang et al., 2012; Sowman et al., 2012; den Ouden et al., 2013; Whymbs et al., 2013). The findings of these studies are very contradictory. While two case reports associated stuttered speech with a decreased activation in left inferior frontal regions (Sowman et al., 2012; den Ouden et al., 2013), a group study linked stuttered speech to an increased activation in this region (Jiang et al., 2012). Furthermore, the case in den Ouden et al., (2013) showed overall more activation during fluent than during stuttered speech, whereas in Whymbs et al., (2013) the majority of the significant findings represented an