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Increased motor preparation activity during fluent single word production in DS: A correlate for stuttering frequency and severity

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ABSTRACT

Abnormal speech motor preparation is suggested to be a neural characteristic of stuttering. One of the neurophysiological substrates of motor preparation is the contingent negative variation (CNV). The CNV is an event-related, slow negative potential that occurs between two defined stimuli. Unfortunately, CNV tasks are rarely studied in developmental stuttering (DS). Therefore, the present study aimed to evaluate motor preparation in DS by use of a CNV task. Twenty five adults who stutter (AWS) and 35 fluent speakers (FS) were included. They performed a picture naming task while an electro-encephalogram was recorded. The slope of the CNV was evaluated at frontal, central and parietal electrode sites. In addition, a correlation analysis was performed with stuttering severity and frequency measures.

There was a marked increase in CNV slope in AWS as compared to FS. This increase was observed over the entire scalp with respect to stimulus onset, and only over the right hemisphere with respect to lip movement onset. Moreover, strong positive correlations were found between CNV slope and stuttering frequency and severity. As the CNV is known to reflect the activity in the basal ganglia-thalamo-cortical-network, the present findings confirm an increased activation of this loop during speech motor preparation in stuttering. The more a person stutters, the more neurons of this cortical–subcortical network seem to be activated. Because this increased CNV slope was observed during fluent single word production, it is discussed whether or not this observation refers to a successful compensation strategy.

1. Introduction

Stuttering is a speech disorder primarily characterized by the occurrence of speech blocks, prolongations and/or repetitions of sound or syllables. These may be accompanied by accessory (secondary) behaviours, i.e. behaviours used to escape and/or avoid these speech events (American Speech-Language-Hearing Association, 1999). When the disorder begins in early childhood, it is called developmental stuttering (DS) (Bloodstein and Ratner, 2008; Van Borsel, 2014). One of the neural characteristics of DS is abnormal motor preparation. Motor preparation contains all processing stages in which a phonological word is transferred into concrete, context-specific articulatory motor commands. According to some theories, it includes phonological word encoding as well (Peters et al., 2000; Indefrey and Levelt, 2004; Indefrey, 2011).

Apart from theoretical arguments for speech motor planning deficits in stuttering (Venkatagiri, 2004; Peters et al., 2000; Packman et al., 1996), neurological evidence is available. The most important cortical structure for motor preparation is the premotor cortex (PMC) with a distinct role of its ventral part (vPMC) for speech (Golfinopoulus et al., 2010). Adjacent to and partly overlapping with vPMC is the inferior frontal gyrus (IFG) which includes in the left hemisphere, the well-known Broca's area (Brodmann area 44, 45). Several studies on stuttering reported both anatomical and functional disturbances in this region. Uni- and bilateral hypo- and hyperactivations have been described at rest, in silent reading and overt speech production (Fox et al.,

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1996; De Nil et al., 2000, 2003; Watkins et al., 2008; Xuan et al., 2012). The most recurrent finding is an anomalous right laterality in activity of the frontal operculum, the homologue of Broca's area (for a meta-analysis, see Brown et al. (2005)). Three magnetoencephalography studies revealed some interesting findings as well. Walla et al. (2004) observed in adults who stutter (AWS) a decreased preparatory activity in or close to bilateral motor cortex preceding overt word reading. Sowman et al. (2012) showed large differences in inferior frontal areas between fluent and stuttered speech. In this case report, blocks, as compared to fluent utterances, were associated with decreased activation in left and increased activation in right IFG extending into orbitofrontal areas. Finally, Salmelin et al. (2000) found an advanced activation of left motor cortex and a delayed activation of left IFG during overt reading. AWS were suggested to initiate motor programmes before preparing the articulatory code. This timing deficit has been linked with decreased white matter density in tracts connecting Broca's area and left motor cortex (Sommer et al., 2002; Chang et al., 2011).

When considering motor preparation, subcortical influences must be taken into account as well. The GODIVA (Gradient Order Directions Into Velocities of Articulators) model, an extension of the DIVA model (Guenther, 2006) provides an explanation on how speech movements are selected, sequenced and initiated (Bohland et al., 2010). This model highlights the crucial role of the thalamus and basal ganglia in motor preparation. These subcortical structures form a reciprocal loop with vPMC: the basal ganglia-thalamo-cortical (BGTC) loop. An alteration of activation in this loop has repeatedly been shown in AWS. Moreover, these altered activations seem to correlate positively with stuttering frequency and severity (Braun et al., 1997; Fox et al., 2000; Giraud et al., 2008; Chang et al., 2009; Kell et al., 2009; Ingham et al., 2012).

One of the electrophysiological substrates of motor preparation is the contingent negative variation (CNV). The CNV is an event-related, slow negative potential that occurs between two defined stimuli. The first stimulus is the warning stimulus (S1) which announces the imperative stimulus (S2) which in its turn requires a response (Walter et al., 1964; Rohrbaugh and Gaillard, 1983; McCallum, 1988; Regan, 1989; Golob et al., 2005). This response is typically a motor response, though cognitive tasks have been reported as well (e.g. Cui et al., 2000; Bares et al., 2007). If the interval between the onset of S1 and S2 is larger than 2 s, two CNVs can be distinguished within this interstimulus interval. The first one, the initial CNV, is induced by and related to orientation to the warning stimulus. It has its largest amplitude at frontal sites within the first second following S1. The second one, the late CNV, occurs before S2 and has a wide cortical distribution with a centro-posterior maximum (Walter et al., 1964; Loveless and Sanford, 1974; Rohrbaugh and Gaillard, 1983; McCallum, 1988; Regan, 1989). The late CNV is reported to have multiple cortical and subcortical generators: prefrontal, premotor, primary motor, anterior cingulate, somatosensory and parietal regions as well as the basal ganglia and thalamus. Hence, the late CNV is generally accepted to measure the neuronal 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). This late CNV is suggested to represent primarily motor preparation, and, additionally, sensory anticipation for S2 (Bender et al., 2004; Bares et al., 2007).

CNV research usually requires a motor response from the limbs. Only a few studies required speech or a non-speech oral movement (e.g. Michalewski and Weinberg, 1977; Yoshida and Iizuka, 2005; Mock et al., 2011). There are even fewer reports concerning stuttering. Those that can be found are dated and have poor methodology according to modern day standards (Zimmerman and Knott, 1974; Pinsky and McAdam, 1980). Prescott and Andrews (1984), and Prescott (1988) evaluated the influence of the complexity of the speech response on the CNV amplitude in AWS. In the former study, AWS displayed larger CNV amplitudes than fluent speakers (FS) but not significantly so. In the latter study, a significant increase was found, but only for familiar words. As familiar words are highly practiced speech responses and therefore very likely to be completely pre-programmed, the authors concluded that AWS have difficulties establishing efficient motor programmes. This concurs with the suggestion of Venkatagiri (2004) that speech motor planning deficits in stuttering may be restricted to familiar syllable motor plans as opposed to new or unfamiliar utterance plans. The above studies mainly focused on the effect of task complexity on motor preparation, and as such the effect of individual variation as to stuttering severity remained unexplored. Interestingly, Zimmerman and Knott (1974) observed large inter-individual variations among stuttering participants.

Recently, we explored the effect of stuttering frequency/severity in a case of acquired stuttering following stroke in left superior temporal gyrus (STG) and stroke related surgery (Vanhoutte et al., 2014). A speech related CNV task involving picture naming was administered at four points in time with differences in stuttering frequency. Late CNV amplitude appeared to be inversely proportional to stuttering frequency during conversation, i.e. the larger the stuttering frequency, the smaller the CNV amplitude which was opposite to the postulated hypothesis. As on the one hand, mostly positive correlations have been described between stuttering severity/frequency and the activity in 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) and on the other hand, the CNV amplitude is known to represent the amount of activity in this loop (Lamarche et al., 1995; Hamano et al., 1997; Gomez et al., 2003; Bares et al., 2007; Fan et al., 2007), an increased amplitude with increasing stuttering frequency was expected. However, previous studies all concerned DS. We hypothesised that, in this patient, the neural network involved in fluent (and stuttered) speech was disturbed differently compared to DS, causing the opposite observation. In DS, the hypothesised lesion site is suggested to be in the proximity of the left IFG (Sommer et al., 2002; Chang et al., 2008, 2011; Watkins et al., 2008; Kell et al., 2009; Cykowski et al., 2010), while in our case study the lesion was situated in left STG. Both regions will cause aberrant auditory–motor integration and will have an adverse effect on the cortical input to the basal ganglia (Giraud et al., 2008). However, the effect may be different because the primary lesion site is different.

Therefore, the present study aimed at evaluating the late CNV as an index of motor preparation activity during overt speech production in AWS with DS. Interactions between structures in the BGTC-loop are complex. In general, however, several structures have repeatedly been reported to show increased activations in stuttering (e.g. Ingham et al., 2012). As such, AWS are hypothesised to show an enlarged CNV amplitude. Secondly, the influence of stuttering severity and frequency was explored by a correlation analysis. In case of a correlation, a positive correlation was expected.

2. Method

2.1. Participants

Originally, 35 AWS with DS and 41 FS were included in the study. Some participants had to be excluded because of (A) technical problems with the microphone ($n=2$), (B) abundant EEG artefacts due to speech ($n=3$), sweating ($n=3$), masseter EMG ($n=1$) or secondary behaviour ($n=2$), and (C) no (pure) DS ($n=2$). Although 28 AWS remained with good quality EEG data, 3 more participants were not included in further analyses because they