

Prosodic processing in people who do and do not stutter: Evidence from pause perception

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Abbreviations

PWS – people who stutter

PWDS – people who do not stutter

AWS – adults who stutter

AWDS – adults who do not stutter

CWS – children who stutter

CWDS – children who do not stutter

EEG – electroencephalography

ERP – event-related potential

fMRI – functional magnetic resonance imaging

TOI – time of interest

ROI – region of interest

CPS – closure positive shift

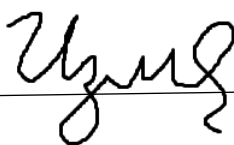
Abstract

Stuttering is a neurodevelopmental disorder characterised by frequent repetitions, prolongations, and pauses. Although stuttering is commonly viewed as a speech production disorder, recent evidence suggests that speech perception may also be affected, e.g. people who stutter (PWS) have been shown to process auditorily presented words and sentences differently to people who do not stutter (PWDS). According to the motor theory of speech perception, speech production mechanisms may in part determine the way speech is perceived. Since PWS' speech production is impaired, it is possible that their perception of speech could be too. Recently, PWS have been found to have disrupted rhythm perception which has been attributed to aberrant functioning in neural timing networks. Given that speech prosody is comprised of speech rhythm and intonation then it might be expected that PWS exhibit prosody processing abnormalities. The aim of this project was to investigate whether PWS processed prosodic information differently to PWDS. To this end, prosodically expected grammatical pauses located at clause boundaries and prosodically unexpected ungrammatical pauses located within syntactic phrases were presented to a group of PWS and an age-matched group of PWDS while their electroencephalogram (EEG) was recorded. Since existing data on neurophysiological correlates of pause perception was scarce, the first part of the project (Experiment 1) was dedicated to identifying neural correlates of grammatical and ungrammatical pause perception in a non-stuttering population. 40 participants listened to sentences that contained grammatical and ungrammatical pauses and watched an unrelated video clip. Event-related analysis of the EEG showed two time windows, -100-100 ms and 100-230 ms relative to pause onset, where the event-related potential (ERP) amplitude difference between grammatical and ungrammatical pauses was statistically significant. In the early time window, ungrammatical pauses were associated with a significantly larger positivity than grammatical pauses. This was unexpected as processing

of phrase boundaries is typically marked with a large slow potential, the closure positive shift (CPS). In the late time window, ungrammatical pauses elicited an enhanced N1 response which was interpreted as a marker of prosodic violation processing. The time windows and regions of statistically significant difference were used to define time and regions of interest (TOI/ROIs) for Experiment 2. Experiment 2 compared ERPs elicited by grammatical and ungrammatical pauses in PWS and PWDS. Participants were 15 PWS and 15 age- and sex-matched PWDS. The experimental design was identical to that of Experiment 1. A 2x2 mixed factorial ANOVA test was performed on the mean ERP amplitude for each TOI using pauses (grammatical, ungrammatical) and fluency (stuttering, non-stuttering) as factors. The results showed that there was no significant difference in ERPs between stuttering and non-stuttering subjects in the early time interval. In the late time interval, a significant interaction between pauses and fluency was found. Stuttering participants produced a significantly reduced N1 response to ungrammatical pauses than non-stutterers. Responses to grammatical pauses were similar across both groups. This finding was interpreted as a diminished ability (at least at the neural level) in PWS to discriminate between expected and unexpected prosodic events during early stages of speech processing and attributed to partial failure to successfully predict prosodic elements in speech. Further studies are needed to determine the behavioural significance of these findings.

Declaration

I declare that this work, either in part or in whole, has not been submitted elsewhere as either part of a degree or for publication, in any other university or institution. The sources of information for this work were wholly from my own research, except when referenced. The writing of this thesis was independent and free from plagiarism. The research was approved by the Macquarie University Ethics Committee (protocol number: 5201400585).

A handwritten signature in black ink, appearing to read 'Tatiana', is written over a horizontal line.

Tatiana Izmaylova

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Chapter 1: Introduction

Stuttering is a speech disorder characterised by frequent pauses, prolongations, and repetitions that affects approximately 5% children and 1% adults (Yairi & Seery, 2015). Stuttering is a very versatile disorder. Despite having being studied for over a century, stuttering is still poorly understood. When people think of stuttering, they usually imagine a person who struggles to speak. This is true. Difficulties with speech production is the most obvious characteristic of stuttering. Yet, no matter how surprising it may sound, stuttering may also involve perception problems. Recent studies have shown that people who stutter (PWS) may process speech differently from people who do not stutter (PWDS; Usler & Weber-Fox, 2015; Weber-Fox, Hampton Wray, & Arnold, 2013). While semantic and syntactic aspects of speech processing have been touched upon in other studies, to date, there have been no neurophysiological studies that examined prosodic processing in PWS.

In this chapter, I will argue why PWS may process speech prosody differently to PWDS. I will start off with providing a historical overview of stuttering theories, including the theory of internal timing deficits in PWS. Then, I will discuss how timing deficits manifest in speech production, how they may affect speech rhythm perception, and what implications disrupted rhythm perception may have for prosodic processing. The role of pauses in prosodic processing will be discussed together with the findings of electrophysiological studies of pauses. Then, the rationale for the present study will be provided.

1.1 Theories of stuttering: A historical overview

Stuttering has been known to the humanity for a very long time. Laozi mentions stuttering in a poem written 2500 years ago: “The greatest wisdom seems like stupidity. The greatest

eloquence like stuttering” (Van Riper, 1982). Ancient Egyptians had a hieroglyph denoting stuttering ‘nitit’ that was used around 2000 BC:



Picture 1. The determinative for the word ‘stuttering’ is a man pointing to his mouth. From Faulkner (2002).

Many famous historical figures stuttered: Virgil, Erasmus, Ludwig the Stutterer, Charles Darwin, King Charles I, just to name a few.

Throughout centuries people held various beliefs about the nature of stuttering. Hippocrates linked stuttering to chronic diarrhoea. Demosthenes was thought to cure his stuttering by practising speaking with pebbles in his mouth. In the 1800s, special devices were worn over the tongue to increase its weight, with the aim to treat stuttering. In France, ivory or golden forks were attached to the lower jaw to support the ‘weak tongue’ (Yairi & Seery, 2015). However, it was not until the 20th century that stuttering received systematic scientific attention. Various theories of stuttering have emerged in the last 100 years. Yet, despite all the advances in science, there is still no theory that provides an exhaustive account of what stuttering actually is.

The 20th century was particularly fruitful in generating numerous theories of stuttering. Some of those theories have been discarded while others have persisted for several decades. In the early 1900’s, theories of stuttering were heavily influenced by psychoanalysis and were based on Freud’s model of child psychosexual development. Most of them viewed stuttering as a deep-seated neurosis that involved a conversion reaction. For example, Coriat (1928) considered stuttering as a fixation at the oral stage of development representing an unconscious conflict between a wish for infantile suckling and a need for an appropriate

behaviour. In parallel to psychoanalytic theories, following Pavlov's work on classical conditioning and Skinner's work on operant conditioning, psychobehavioural theories of stuttering began to emerge. Stuttering was viewed as a learnt behaviour that at some point developed into a habit. Johnson (1942) proposed that children developed stuttering due to their parents' negative reaction to normal speech disfluencies that occurred during language acquisition. As a result, children became anxious and attempted to avoid disfluencies which lead to a built-up tension that, over time, developed into stuttering. From this point of view, stuttering was nothing but an anticipatory avoidance behaviour. Along the lines of the behavioural approach, the demands-capacities model of stuttering was formed (Adams, 1990). The model posits that stuttering occurs when a child attempts speech that is beyond his or her current capabilities. According to this model, stuttering resolves when the child's capacities meet or surpass speech demands.

Another very popular theory that has not been supported by later research is the theory of cerebral dominance proposed by Lee Travis (1931). In accordance with this model, in order to produce speech movement patterns, both cerebral hemispheres should operate in a synchronised manner, precisely, one hemisphere should lead in movement initiation and the other should follow it. However, PWS lack cerebral dominance which results in both hemispheres sending signals for movement initiation, thus, creating an asynchrony in neural timing and disruption of speech.

Psycholinguistic theories of stuttering focus on how PWS conceptualise, formulate, and articulate speech. In PWDS these processes are automatic and take place parallelly (Levelt, Roelofs, & Meyer, 1999). This may not be the case with PWS. According to the covert-repair model, PWS detect an upcoming error and attempt to correct it before the error reaches the level of speech production, which results in stuttering. Perkins, Kent, and Curlee (1991) propose a model that stresses importance of synchronised production of syllable frames and

speech sounds that fill them up. They reason that stuttering occurs when either syllable frames or speech sounds are not ready but the time pressure demands that PWS go ahead with the speech.

Stuttering can be viewed as a sensorimotor dysfunction. According to this approach, PWS have difficulties integrating feedforward and feedback motor control. Planning and initiation of speech movement is performed by feedforward motor control. After the movement has been initiated, it can be adjusted through the feedback system. Civier, Tasko, and Guenther (2010) suggest that PWS have impaired feedforward systems and rely exceedingly on the feedback system when producing speech which leads to an increased amount of ‘repair’ information causing the system to ‘reset’ or repeat syllables. Similarly, Zimmermann (1980) suggests that stuttering is a movement disorder caused by imbalances in afferent-efferent interactions of brain-stem structures.

From the auditory perspective, stuttering occurs due to a perceptual auditory defect in PWS. The defective auditory feedback interferes with speech motor activity, causing speech disruptions (Lincoln, Packman, & Onslow, 2006). This perspective is supported by findings that PWS’ speech can be improved when they do not receive auditory feedback, such as speech in noise, or when auditory feedback is altered (delayed or frequency-shifted).

Modern theories of stuttering are predominantly based on studies of the brain physiology and functioning. They attempt to identify the cause of stuttering by comparing brain functional or structural differences between PWS and PWDS. PWS are known to have an increased volume of white matter in the right hemispheric auditory areas and Broca’s area right-sided homologue which suggests abnormal communication within the right hemisphere that may affect speech processing strategies (Jürgen, Lutz, & Helmuth, 2004). Significantly reduced white matter integrity is found in the bilateral arcuate fasciculus, a white matter tract that

connects Broca's and Wernicke's areas and is implicated in speech (Connally, Ward, Howell, & Watkins, 2014). PWS have been found to have a larger corpus callosum which may suggest an increased communication between the cerebral hemispheres, perhaps, due to abnormal distribution of language processing (Choo et al., 2011). Reduced grey matter is found in the caudate nucleus of the basal ganglia in adults who stutter (AWS) suggesting that AWS may have deficient selection and sequencing of speech motor movements (Sowman et al., 2017). Functional magnetic resonance imaging (fMRI) studies report an overactivation of the right inferior frontal gyrus (Broca's right-sided homologue) and underactivation of the bilateral auditory cortex (Brown, Ingham, Ingham, Laird, & Fox, 2005). Like many other theories before, neurophysiological approach has not yet come up with a unifying explanation of stuttering.

While most theories are concerned with aberrant speech production in PWS, some theories suggest that speech perception may also be impaired in stuttering. Motor theory of speech perception posits that speech is perceived through covert speech production. According to this theory, listeners do not perceive phonology and prosody of speech, they perceive a series of vocal gestures. The theory assumes that speech perception involves access to the speech motor system (Galantucci, Fowler, & Turvey, 2006; Liberman & Mattingly, 1985).

According to this theory, PWS may experience deficits in speech perception due to the impaired motor system involved in speech production. Recent neuroimaging studies provide support to this perspective (Halag-Milo et al., 2016; Lu et al., 2016a).

Another attempt to explain stuttering that is not mutually exclusive with the motor theory of speech perception is the theory of internal timing deficits in PWS (Etchell, Johnson, & Sowman, 2014; Van Riper, 1982). According to this theory, PWS have deficits in an internal timing network that cause disruptions in PWS' motor productions, including production of speech. Recent studies suggest that PWS have an impaired rhythm perception, presumably,

due to deficient internal timing (Chang, Chow, Wieland, & McAuley, 2016; Wieland, McAuley, Dilley, & Chang, 2015). According to Harrington (1988), the ability to perceive rhythm is crucial for speech processing. As a result of internal timing deficits and impaired rhythm perception, PWS may experience difficulties with speech processing. This theory is particularly compelling as it proposes a common mechanism that underlies both motor productions and auditory perception in PWS, providing the most parsimonious view on the cause of stuttering.

1.2 Timing deficits and speech production in PWS

One of the theories attempting to explain stuttering posits that stuttering is an internal timing disorder (Etchell, Johnson, & Sowman, 2014; Harrington, 1988; Van Riper, 1982). That theory stems from the idea that stuttered speech is full of temporal irregularities. PWS were found to have longer voice onset times and longer voice termination times even in fluent utterances (Agnello, 1975; Agnello, Wingate, & Wendell, 1974). Cooper and Allen (1977) reported that PWS had a greater variability of durations in a sentence repetition task. Similarly, PWS showed a significantly greater variability in the length of intervals between two consecutive fundamental frequency peaks associated with stressed syllables in reading tasks (Bergmann, 1986). Boutsen, Brutten, and Watts (2000) investigated intensity and timing variability in PWS' speech under a metronome condition that is known to induce fluency in PWS. They found that the intensity variability did not differ between the groups while durations between successive syllable onsets in PWS were significantly more variable. Falk, Maslow, Thum, and Hoole (2016) examined temporal variability in spoken and sung productions of stuttering and fluent adolescents. Their findings showed that although vowel and utterance durational variability reduced in sung productions, there was still a significant difference in utterance durations between stuttering and non-stuttering groups.

It has been known for a long time that PWS become fluent when they synchronise their speech with a stimulus that provides additional timing cues, e.g. pacing speech to a metronome, singing or speaking in unison with another person (Ingham & Carroll, 1977; Van Riper, 1982; Wohl, 1968). According to Etchell et al. (2014) this is because PWS' have deficits in the internal timing network (comprised of the basal ganglia and the supplementary motor area) that becomes active when individuals make self-initiated movement. Due to internal timing deficits, PWS rely on the external timing network (comprised of the cerebellum, the premotor cortex, and the right inferior gyrus) that becomes activated in the presence of external stimuli, which is why PWS respond well to fluency inducing conditions.

Support for the timing deficit theory comes from neurophysiological studies that find differences in neural networks between stuttering and non-stuttering populations. Of particular importance are findings of impaired basal ganglia functioning in PWS. The basal ganglia are known to be implicated in timing of self-generated movement, control of movement sequences, and sensorimotor integration, functions that are crucial for speech production. Chang and Zhu (2013) examined functional and structural connectivity in the neural networks of stuttering and fluent children. They found that both functional and structural connectivity between the putamen and the supplementary motor area, the largest areas within the basal ganglia-thalamocortical network, were attenuated in children who stutter (CWS). Toyomura, Fujii, and Kuriki (2015) reported normalisation of the cerebellum (decrease) and the basal ganglia (increase) activity during speech in PWS after 8 weeks of metronome-paced practice. In sum, speech motor productions of PWS are temporally irregular and the cause of this irregularity has been attributed to internal timing deficits that may result from impaired functioning of the basal ganglia-thalamocortical circuit.

1.3 Rhythm

1.3.1 Speech rhythm

While impaired internal timing affects speech production in PWS, less is known about PWS' speech perception. According to (Harrington, 1988), the ability to predict upcoming speech elements is crucial for successful speech processing. In order to make predictions about speech elements, one should assume that speech bears an element of predictability. This predictability is captured in the notion of speech rhythm. A classic definition of speech rhythm is based on a periodicity assumption that expects repetition of strong elements over equal time intervals (Lowit, 2014). Recent research in this field, however, shows that speech rhythm is much more complex and less predictable than the classic theory suggests (Jadoul, Ravnani, Thompson, Filippi, & de Boer, 2016; Nolan & Jeon, 2014; Turk & Shattuck-Hufnagel, 2014). Jadoul et al. (2016) examined predictability of speech rhythm in 18 languages. They found limited regularity at the level of syllable inter-nucleus durations. That regularity, however, was not sufficient for a subjective experience of rhythm in speech. The authors concluded that rhythm perception may be determined by top-down expectations imposed on weak regularities found in multiple prosodic parameters, such as pitch, segment durations, intensity, and inter-nuclear intervals. Nolan and Jeon (2014) support this view by arguing that salient prosodic prominence (e.g. stress) promotes an analogy with beats in music even in the absence of actual regularity between salient elements. Grahn (2009) suggests that successful rhythm perception requires an element of expectancy. Two conclusions can be made from the above. First, it appears that an ability to form expectations about upcoming elements is crucial for speech rhythm perception. Second, superimposing regularity on not-so-regular prosodic patterns requires well-coordinated bottom-up and top-down processing.

1.3.2 Rhythm perception in PWS

Speech rhythm perception is an important part of speech processing as it allows the listener to identify strong and weak elements in speech and predict an emerging prosodic structure. As was discussed earlier, PWS exhibit degraded rhythmic structure in their motor productions. If we assume that rhythmic production ability has implications for predictive aspects of perception, we might hypothesise that rhythm perception may be impaired in PWS. Indeed, evidence is emerging that PWS have an impaired perception even of relatively simple non-speech rhythms. In the first ever study of rhythm perception in stuttering population, Wieland, McAuley, Dilley, and Chang (2015) examined rhythm discrimination abilities in CWS and children who do not stutter (CWDS). Children were exposed to a simple or a complex rhythm sequence twice, after which they needed to judge whether the third rhythm sequence was the same or different from the previous one. CWS were significantly worse at both simple and complex rhythm discrimination than CWDS.

Impaired performance on production and perception tasks may be explained by overlaps in the neural networks involved in both processes. The same basal ganglia thalamocortical network involved in dysrhythmic speech production in PWS was found to be implicated in rhythm perception. Specifically, Chang, Chow, Wieland, and McAuley (2016) examined correlation between spontaneous brain activity in the rhythm network comprising the putamen, supplementary motor area, premotor cortex, and auditory areas in the bilateral superior temporal gyrus and performance on a rhythm perception task in CWS and CWDS. They found that, compared to controls, functional connectivity between the putamen and the SMA was attenuated in CWS. Additionally, while CWDS showed a strong correlation between intrinsic functional connectivity and performance on the rhythm discrimination task, no such correlation was found in CWS. The results of the study suggest that the basal ganglia

could be the source of internal timing not only for motor productions but also for rhythm perception.

Grahn (2009) examined the role of the basal ganglia in rhythm perception. In an fMRI study, she compared activation of the basal ganglia in response to three types of beat accents: temporal (duration), dynamic (volume), and subjective (no accent). While in the temporal and dynamic conditions, beats were marked with an increased duration or volume, in the subjective beat condition, no accents were present. Participants reported perceiving beat even in the unaccented (subjective) beat condition. Activation of the basal ganglia was highest for the subjective beat condition. The author concluded that the basal ganglia was linked to internal beat generation. She also reported that other components of the basal ganglia-thalamocortical network, such as the premotor cortex, SMA, and the superior temporal gyrus were involved in rhythm processing. The results of this study support the claim that PWS may experience difficulties with rhythm perception due to impaired functioning of the basal ganglia-thalamocortical network. Since speech perception requires superimposition of an internally-generated rhythm onto irregular prosodic patterns which, speculatively, may be supported by the same basal ganglia-thalamocortical network, PWS may show some impairments in speech rhythm perception. This idea is supported by Kotz and Schmidt-Kassow's (2015) study that examined the P600 response to syntactic and metrical structure violations in patients with lesions to the basal ganglia due to stroke. While controls showed a stable P600 response to both types of violations, basal ganglia patients responded only to syntactic violations, failing to differentiate between metrically expected and unexpected speech events. To conclude, it appears that the basal ganglia-thalamocortical network dysfunction may impair an individual's ability to successfully perceive rhythm, as well as build predictions about upcoming prosodic elements, which may lead to difficulties with

online speech processing. If this is the case, speech perception studies should find differences between PWS and PWDS in how they process speech.

1.4 Neurophysiological studies of speech perception in PWS

Studies that have examined speech perception in PWS, suggest that PWS, indeed, process speech differently to PWDS. Most of them point to differences between PWS and PWDS in the late event-related potential (ERP) components related to cognitive processing of information. For example, Weber-Fox (2008) examined event-related potentials (ERPs) to auditorily presented semantic violations represented by an unexpected verb (e.g. “Every day, the children *rust to be superheroes”) and syntactic violations represented by a verb agreement (e.g. “Every day, the children *pretends to be superheroes”) in AWS. The results revealed that, in controls, an N400 was elicited to semantic and P600 to syntactic violations, as expected. In contrast, each condition elicited both an N400 and a P600 in AWS. Additionally, both peaks were reduced in AWS. Appearance of an N400 and a P600 in both conditions was completely unexpected and may have signified that AWS employed semantic-syntactic processing in a more general way compared to AWDS. In another experiment, CWS produced an N400 in response to syntax violation of Jabberwocky sentences as opposed to a P600 that was produced by CWDS and recovered CWS (Usler & Weber-Fox, 2015). The authors rationalised that appearance of the N400 suggested that PWS relied on semantic cues to comprehend syntactic context. As the ability to process speech relying on semantic cues develops earlier in life, the researchers concluded that neural mechanisms associated with syntactic processing could be less mature in CWS compared to CWDS.

Apart from the N400 and P600, early ERP components related to sensory processing have also been found to be abnormal in stuttering. (Weber-Fox et al., 2013) studied ERPs to auditorily presented semantic and phrase structure violations in pre-school CWS and CWDS.

They reported that, compared to CWDS, CWS showed an increased early negativity between 150-350 ms after the stimulus onset over medial electrodes. This early negativity is commonly associated with online syntactic parsing and building a local phrase structure. The N400 peak latencies were found to be longer in CWS and the P600 amplitude was larger over the right hemisphere in CWS and over the left hemisphere in CWDS. These findings were interpreted as an example of developmental differences between preschool PWS and PWDS, as well as an indicator of an early atypical hemispheric lateralisation that is usually observed in AWS.

Differences between PWS and PWDS have been found in early ERPs using an oddball paradigm. Corbera, Corral, Escera, and Idiazabal (2005) investigated mismatch-negativity (MMN) responses to pure tone and phonetic contrasts in PWS and PWDS. Their findings revealed no differences between the two groups in pure tone perception, however, PWS produced enhanced supratemporal left-lateralised MMN responses to phonetic contrasts compared to PWDS. The authors attributed these findings to abnormal memory traces for speech sound representations in PWS and suggested that it could be an underlying cause of the disorder.

In a similar magnetoencephalographic (MEG) study, Biermann-Ruben, Salmelin, and Schnitzler (2005) examined neural activation in PWS and PWDS during exposure to pure tones, words that they had to repeat and sentences that they had to either repeat or transform into passive forms. The authors found that there were no differences between the two groups on the non-speech task. During exposure to speech, however, two additional areas were activated in PWS: the left inferior frontal cortex and the right rolandic area. The authors interpreted an increased activation of the left inferior frontal cortex as anticipation of an enlarged load of articulatory planning. Activation of the right rolandic area was attributed to the atypical cerebral dominance which is found during speech tasks in PWS.

fMRI studies that have investigated neural correlates of speech perception in PWS and PWDS have also reported differences between the two groups. Halag-Milo et al. (2016) investigated brain activation during speech perception in PWS and PWDS. An fMRI scan was performed while participants were listening to several short poems. The results revealed that PWS had a stronger activation in the right inferior frontal gyrus and left Heschl's gyrus. As the right inferior frontal gyrus is commonly activated during speech production in PWS, the authors concluded that a single imbalanced network was the cause of speech perception and production deficits in PWS. In a word perception task, De Nil et al. (2008) found an increased activation of the left middle and superior temporal gyri and right insula, primary and supplementary motor cortex in PWS compared to PWDS. The finding was interpreted as an evidence of a more sequential approach to processing words in PWS.

In sum, neurophysiological studies confirm that PWS process speech in a systematically different way from PWDS. One interpretation which could incorporate all these findings is that due to a decreased ability to perceive speech rhythm, PWS may process speech less efficiently compared to PWDS, as can be seen from additional ERP components, higher ERP amplitudes, and increased activation of cortical and subcortical areas in PWS.

1.5 Do PWS process prosodic information differently?

Studies of speech perception show that PWS process speech differently to PWDS. While most studies to date have focused on syntactic-semantic, single word or pure tone processing, no neurophysiological studies have examined whether PWS and PWDS process prosody differently. Prosody comprises speech rhythm and intonation. As PWS experience difficulties with rhythm perception in general (Chang et al., 2016; Wieland et al., 2015), they are likely to have problems with perceiving speech rhythm, which is crucial for prosodic processing. Therefore, it is possible that prosodic processing in PWS may also be impaired.

It has been shown that the prosodic-syntactic interaction occurs early in speech processing (Eckstein & Friederici, 2006). This means that prosodic deficits may interfere with construction of the initial syntactic structure, that occurs before lexical-semantic information is accessed during speech processing (Friederici, 2002). As a consequence, PWS may need to reanalyse syntactic structures more often than PWDS which may result in additional P600 components similar to those observed in response semantic violations (as opposed to only N400 in non-stutterers; Hampton & Weber-Fox, 2008). Impaired prosodic-syntactic processing may also create a tendency to rely more on semantic information, as reported by Usler and Weber-Fox (2015). Although indirectly, the results of the mentioned above studies suggest that prosody may be implicated in disruptions of syntactic processing. Thus, learning more about prosodic processing in PWS would contribute to our understanding of how PWS process speech.

Pauses are prosodic breaks that may assist or disrupt syntactic processing. One of the most important functions of pauses in speech is marking syntactic boundaries. Young children predominantly rely on pauses for processing prosodic phrases (Männel & Friederici, 2011). Pauses help to disambiguate locally ambiguous sentences prior to the onset of the critical word (e.g. “The reporter interviewed the squatter and the policeman in front of the statue in the centre of the city”; Kerkhofs, Vonk, Schriefers, & Chwilla, 2008). At the same time, unexpected pauses disrupt processing of disambiguating words if those are preceded by a pause (Maxfield, Lyon, & Silliman, 2009). The difference between predictable pauses that assist syntactic processing and unpredictable pauses that disrupt it should be very obvious. However, perceiving differences between prosodically expected and unexpected events can be challenging for people who may have difficulties with prosodic processing. As such, pauses may be the ideal substrate for examining prosodic disturbance.

1.6 Pauses in speech

Broadly, pauses are periods of silence in speech. Pauses are important for speech production and speech comprehension as they allow the speaker to formulate their thoughts and provide information about underlying syntactic structures to the listener (Goldman-Eisler, 1972; Reich, 1980). There is another type of pauses, namely, filled pauses, that occur in places of hesitation or utterance formulation (Rochester, 1973). As only silent pauses are reflective of sentence syntactic structure and can be predicted from the context, filled pauses will not be further discussed in this work.

Despite the fact, that research on pauses has accumulated a lot of data since its start in the early 1950's, there is no systematic classification of silent pauses. Below, is an attempt to group silent pauses into the three categories commonly described in the literature on pauses: length, position in a sentence, and grammatical status.

1.7 Pause classification

1.7.1 By length

One of the most basic parameters that can be used for pause classification is pause length (duration). Goldman-Eisler (1961) was one of the first researchers who studied the connection between pause lengths and their distribution in speech. In her classic study, she investigated the length and distribution of pauses in four spontaneous speech samples: a cartoon description, a cartoon summary, a discussion, and a psychiatric interview. She found that pauses of less than 1 s duration were more frequent than pauses of any other length. Pauses were never longer than 3 s and 99% were shorter than 2 s in the discussion task, while the cartoon description, summary, and psychiatric interview contained longer-than-3-s pauses. Goldman-Eisler (1961) did not propose a well-defined classification, however she

suggested that pauses less than 0.25 s long should be viewed as articulatory, while pauses longer than 0.25 s could be attributed to hesitation. Many subsequent studies adopted the same approach and defined hesitation-related pauses as gaps in speech lasting longer than 0.25 s (De Jong, 2016; Boomer & Dittmann, 1962).

In a more recent study, Campione and Veronis (2002) posited the existence of three pause duration categories while investigating a distribution of silent pauses across five languages: brief (below 200 ms), medium (200-1000 ms), and long (over 1000 ms). The authors reported that pauses shorter than 200 ms make up 3.9% of all pauses in the English language and 17.9% in the Italian language. Medium length pauses with a peak duration of around 500 ms are the most common in spontaneous and read speech across the languages.

1.7.2 By position

Pauses can be classified into two categories based on the position of their occurrence within a sentence: 1) pauses that occur between clauses that make up a sentence and 2) pauses that occur within clauses. Pauses that occur within clauses usually reflect difficulties with lexical retrieval of relatively unpredictable words. Pauses between clauses are associated with a more general planning of subsequent syntactic units (e.g. word order, syntactic encoding; Maclay and Osgood, 1959).

Neuroimaging research supports this distinction between two types of pauses. Kircher, Brammer, Levelt, Bartels, and McGuire (2004) performed fMRI scans when participants were describing seven Rorschach inkblots. Pauses between 550 - 3000 ms were selected as events of interest. The authors of this study found that pauses between clauses had a longer mean duration (1277 ms vs. 1037 ms.) and were followed by higher frequency words than pauses within clauses. Pauses within clauses were associated with activation in the left superior temporal, superior frontal, as well as in the middle temporal and middle frontal gyri

bilaterally. Pauses between clauses were associated with activation in the right inferior frontal gyrus. The brain areas implicated in within-clause pause production (the bilateral superior and middle temporal gyri) were previously found to be associated with lexical retrieval and error correction (McGuire, Silbersweig, & Frith, 1996).

Differences between pauses within and between clauses have been found in second language research. De Jong (2016) found no difference in the number of between-utterance pauses between first and second-language speakers of Dutch. However, second-language speakers produced twice the number of within-clause pauses compared to the first-language speakers, presumably, due to the fact that they had to make more frequent lexical decisions than native speakers. These findings support the idea that within-clause pauses are associated with lexical retrieval while between-clause pauses reflect syntactic planning.

1.7.3 By grammaticality

Grammatical pauses are defined as pauses that occur at major syntactic boundaries, while ungrammatical pauses are found within minor syntactic phrases (Ramanarayanan, Bresch, Byrd, Goldstein, & Narayanan, 2009). This definition does not specify, however, what exactly constitutes minor syntactic phrases, which leaves the definition of ungrammatical pauses open to interpretation. As a result, those studies that have investigated grammaticality differences in pausing behaviour have created their own definitions of grammatical and ungrammatical pauses. For example, in their analysis of pauses in spontaneous speech using real-time magnetic resonance imaging, Ramanarayanan et al. (2009) adopted a definition of grammatical pauses in terms of their position within syntactic units. They specified that only pauses between clauses/sentences, pauses at subject-verb and verb-object boundaries, and pauses after prepositional phrases were considered grammatical for the purpose of their study. Another approach to defining grammatical and ungrammatical pauses is based on

where pause occurs in relation to content and function words. Pauses that occur between content words are always considered grammatical, pauses between a function word and a content words are always considered ungrammatical, while the content-function word and function-function word positions are considered grammatical only if they occur between phrases (Beltrame et al., 2011; Thurber & Tager-Flusberg, 1993). Overall, the grammaticality status of pauses appears to be a somewhat subjective characteristic. It is plausible that pause grammaticality judgement is more of a continuum than a binary decision, with some pause locations in a sentence being perceived as more grammatical than others.

Grammatical and ungrammatical pauses tend to differ in their durations. Comparisons of the two reveal that grammatical pauses have a tendency to be longer than ungrammatical ones. There are differences in how prepausal syllables are realised before grammatical and ungrammatical pauses. 70% grammatical pauses are accompanied by a rise in fundamental frequency in prepausal words, while this occurs only for a few ungrammatical pauses (O'Shaughnessy, 1992). Ramanarayanan et al. (2009) reported a large drop in articulatory speed before grammatical pauses, but only a small decrease in the articulatory speed prior to ungrammatical pauses, thence followed by a rapid increase in articulatory speed after the pause.

1.7.4 Classification overlaps

There are a number of overlaps in pause classification as different studies have used different terms to describe the same phenomena. For example, while some studies discuss grammatical and ungrammatical pauses, others refer to them as syntactic junctures and hesitation pauses. The situation becomes more complicated when short articulatory pauses are contrasted with hesitation pauses, in which case it is unclear whether hesitation pauses include both grammatical and ungrammatical pauses, or ungrammatical pauses only.

For the purpose of the current study, it will be assumed that pauses between clauses in complex sentences are grammatical, while pauses occurring within clauses are ungrammatical. Of the ungrammatical pauses, pauses between a function and a content word in a noun phrase will be considered the lowest on the grammaticality spectrum. Pauses between other constituents of a clause (e.g. subject + predicate or transitive verb + object) will still be considered ungrammatical but higher in their grammaticality status. This information will be used as a guiding principle for the experiment design of the present study.

1.8 Electrophysiological studies of pauses

The first electroencephalographic (EEG) study of pauses in speech, to my knowledge, was performed by Besson, Faita, Czternasty, and Kutas (1997). These authors measured ERP responses to 600-ms silent pauses introduced before two final words in sentences. Their results revealed a large evoked potential at the time where the final words should have been presented. The evoked potential comprised a negative component 200 ms after the introduction of silence, followed by a large positive component at around 350-400 ms. After introduction of the final words, an N1-P2 complex was observed followed by an N400 component which the authors associated with semantic integration of words into the sentence context. There was no evoked potential for the no-delay condition, but a slow positive shift was observed from 600 ms to the end of the recording epoch. Besson et al. did not specify what type of pauses they used in their study. Speculatively, it is likely that the pauses were ungrammatical as 2-word clauses at the end of sentences would be unlikely to occur in typical grammatical structures.

Some studies have investigated how silent pauses affect subsequent processing of speech. MacGregor, Corley, and Donaldson (2010) looked into how pauses before predictable and unpredictable sentence endings affected their lexical integration. They found that the

difference in the N400 effect between predictable and unpredictable words was attenuated when they were preceded by silent pauses. Unpredictable words produced a late frontal positivity when they followed silent pauses. The subsequent memory test revealed that participants better remembered unpredictable words, as well as words preceded by silent pauses. Thus, it appears that pauses may facilitate memory retention. Another study reported a similar reduction in the difference of the N400 effect between ‘yes’ and ‘no’ responses when they were preceded by a 1000 ms pause (as opposed to a short pause condition; Bögels, Kendrick, & Levinson, 2015). Kerkhofs et al. (2008) found that pauses at syntactic boundaries helped to disambiguate “garden-path” sentences (i.e. sentences that can be interpreted in two different ways due to their syntactic structures) prior to the lexical disambiguation point.

Other EEG studies have focused on grammatical pauses that mark syntactic boundaries. In their seminal paper, Steinhauer et al. (1999) examined neural correlates of prosodic boundary processing. They measured ERPs in subjects exposed to sentences with one or two prosodic breaks. It was found that the number of prosodic breaks in a sentence corresponded to the number of positive shifts in subjects’ ERPs. The shift was even present in artificially manipulated sentences where prosodic boundaries did not correspond to syntactic boundaries. The authors speculated that the shift was elicited by the intonation of closure commonly present at syntactic boundaries and termed it the closure positive shift (CPS).

Following Steinhauer’s et al. (1999) study, several papers dedicated to pauses between clauses and the CPS were published generally agreeing in their findings of CPS in response to prosodic breaks. Männel, Schipke, and Friederici (2013) argued that pauses play an important role in speech acquisition. They studied ERPs of 3- and 6-year-old children who listened to sentences that did and did not contain pauses at prosodic boundaries. While 6-year-olds showed the CPS in response to both types of sentences, 3-year-olds did so only in

response to the sentences with prosodic breaks. The authors concluded that during the speech acquisition phase, children need pauses in addition to other prosodic boundary markers (pitch rise and preboundary vowel lengthening) to infer phrase boundaries. After they develop more expertise in the language, they cease relying on pauses in their syntactic analysis. In a follow-up study, Männel and Friederici (2016) measured ERPs of 3-year-old children in response to prosodic boundaries that were marked with pauses and preboundary lengthening but were pitch-neutralised. They found that even partially marked prosodic boundaries elicited the CPS when pauses were present. Interestingly, it appears that the CPS may be a universal phenomenon. Li and Yang (2009) reported the CPS in response to intonational phrase boundaries as well as phonological phrase boundaries in Chinese (however, see Strelnikov, Vorobyev, Chernigovskaya, and Medvedev (2006) for different results).

In sum, it appears that pauses between clauses (grammatical pauses) and pauses within syntactic structures (ungrammatical pauses) elicit different neural responses. While there is a consensus on the fact that silent pauses at syntactic boundaries produce the CPS, the neural correlates of pauses within syntactic structures are not so well established due to a paucity of research into this phenomenon. Of the two studies that investigated ERP responses to silent pauses within clauses, only one reported on neural correlates of pauses per se, while the other one focused on the effect of pauses on processing of subsequent words. Additionally, the studies that employed pauses within clauses did not clarify their grammatical status. While it is possible to speculate that a large percentage of them could be ungrammatical, more information on their grammatical status would be desirable. Examining ERP responses to grammatical and ungrammatical pauses consistently would allow direct comparison of differential ERP elicitation by these different prosodic events.

1.9 Pausing behaviour of PWS

There are two types of pauses that occur in the speech of PWS: pauses that constitute a type of disfluency and pauses that are part of fluent speech. Disfluent pauses are caused by an arrest of speech and are called blocks (Yairi & Seery, 2015). Blocks are characterised by a complete closure of PWS' airways and are accompanied by strong increases in the interoral air pressure (Hutchinson, 1975). Griggs and Still (1979) opined that blocks could be related to clause boundary pauses. They discovered that those PWS who produced the largest number of blocks also tended to stutter more on sentence initial words and function words (as opposed to more common stuttering on content words). These authors suggested that silent blocks could be prolongations of natural pauses that commonly occur before function words within and between sentences. The idea that the occurrence of silent blocks could be mediated by the number of naturally occurring pauses is very insightful as most studies do not differentiate between types of dysfluencies when reporting stuttering loci. However, the overall tendency for stuttering events to fall on the beginning of sentences or clauses is well documented in stuttering literature (Van Riper, 1982; Wingate, 1988; Yari & Seery, 2015). According to Wall, Starkweather, and Cairns (1981), stuttering occurs on 28% clause-initial words as opposed to 4% words found within clauses. Interestingly, despite the apparent similarity between blocks and other stuttering events, blocks are most negatively perceived by PWS who react to the temporal inability to initiate speech with "panic and frustration" (Van Riper, 1982, p. 122).

In addition to blocks, the pausing behaviour of PWS differs from that of PWDS even when their speech is fluent (Wingate, 1984). One of the first studies of PWS' and PWDS' pausing patterns by Love and Jeffress (1971) found a significantly larger number of short pauses (150-250 ms duration) in PWS' speech. More than 10 years later, Winkler and Ramig (1986)

examined the number and duration of pauses in the speech of CWS and CWDS on simple (repeating sentences) and complex (retelling short stories) speech tasks. They found that on complex speech tasks, CWS produced a significantly larger number of pauses than CWDS did. They also reported that interword pause duration was significantly longer in CWS than CWDS on both types of tasks. Their results demonstrate not only that PWS and PWDS differ in the number of pauses they produce, but also that the syntactic structure of text can affect stuttering and non-stuttering groups differently.

Similarly, Yoshiyuki and Ramig (1987) compared the duration of pauses in the first and the sixth reading of the Rainbow Passage by PWS and PWDS. They established that PWS had significantly longer mean pause durations compared to PWDS. Although the authors did not discuss syntactic complexity of the task, it is known that the Rainbow Passage contains many compound and complex sentences which could differentially affect the length of pauses produced by PWS and PWDS. Subsequent research confirmed that syntactic boundaries may, indeed, be processed differently by PWS. Klouda and Cooper (1987) reported that severe stutterers did not pause consistently at syntactic boundaries while less severe stutterers did.

The abovementioned studies provide evidence to the claim that PWS pause more frequently and less consistently even when they are fluent. Syntactic complexity may also play a role in the distribution of pauses, with more pauses occurring in complex sentences. At least, some PWS are inconsistent in their pausing at syntactic boundaries. All of the above suggest that PWS produce highly irregular prosodic patterns interspersed with unexpected prosodic breaks. Following our earlier discussion, if speech production and perception are linked via a common underlying mechanism (such as an internal timing network), it could be expected that PWS' prosody perception may also be impaired.

1.10 The present study

This study set out to investigate whether prosodic processing in PWS was different to that of PWDS. To do this, we decided to investigate responses to pauses as pauses are an integral part of speech prosody and an experimentally manipulatable and controllable constituent of a sentence. As discussed earlier, PWS produce significantly more pauses in their fluent speech compared to PWDS (Wingate, 1984). In the light of the speech timing discussion, one might anticipate that the ability to correctly perceive speech rhythm would play an important role in grouping words into larger constituents and assigning correct prosodic patterns to them during speech production. As it is contended that PWS have internal timing deficits, likely, due to a basal ganglia dysfunction, they might be expected to experience difficulties in predicting where pauses should be placed in order to preserve prosodically correct structures. Since speech perception might be affected by deficits in the functioning of the internal timing network, PWS may have difficulties with processing speech prosody. If this is true, then PWS may process expected pauses at syntactic boundaries and unexpected pauses within syntactic units differently to PWDS. For convenience, unexpected and expected pauses will be referred to as ungrammatical and grammatical respectively.

The aim of this study was to investigate neural responses to grammatical and ungrammatical pauses in PWS and PWDS. For this purpose, a passive ERP paradigm was chosen. The ERP method is characterised by high temporal resolution which is ideal for examining online speech processing. No control for attention was implemented as it would require concomitant verbal or non-verbal responses to assess. The requirement for overt responses can be problematic for stuttering studies as motor response preparation in PWS is associated with abnormal neural activity (e.g. overactivation of the right rolandic areas during speech preparation; Biermann-Ruben et al., 2005), which could confound the observation of any

aberrant brain activity during speech perception. It is important to point out that an ERP study would only provide information about neural correlates of prosodically expected and unexpected events, which may or may not be reflective of the actual perception of prosody by PWS and may or may not explicitly show up at the behavioural level.

Prior to investigating neural processing of pauses in PWS, it was important to establish whether there were differences between neural responses to grammatical and ungrammatical pauses in non-stutterers. While grammatical pauses are often discussed in the CPS studies (Männel & Friederici, 2011; Männel et al., 2013; Steinhauer et al., 1999), ERPs to grammatical pauses per se (not in relation to the CPS) have never been examined. Our understanding of ungrammatical pauses is even more limited. The only study that explored ERP responses to pauses inserted before the two sentence-final words found a large negativity peaking 200 ms after the pause onset followed by a positive component peaking around 350-400 ms. The grammaticality status of pauses, however, was not clearly stated (Besson et al., 1997). Due to a paucity of information on the topic, a preliminary study of pause perception in a non-stuttering population was needed.

The present study consisted of two separate experiments. The aim of Experiment 1 was to establish neural correlates of grammatical and ungrammatical pause perception in PWDS. The results of Experiment 1 were interpreted from the perspective of predictive processing and used for designing Experiment 2. In particular, time windows and electrodes where significance between grammatical and ungrammatical pauses was found were used as time and regions of interest (TOI/ROIs) for Experiment 2. The aim of Experiment 2 was to find out whether PWS process prosodically expected and unexpected events (represented by grammatical and ungrammatical pauses) differently from PWDS.

Chapter 2: Neural correlates of grammatical and ungrammatical pause perception in PWDS (Experiment 1)

2.1 Hypothesis

Experiment 1 was a preliminary study for Experiment 2. Its purpose was to acquire normative physiological data on pause processing in a non-stuttering population that could be used as a priori constraints for subsequent investigations in a stuttering population. The results of Experiment 1 were used in formulating the hypothesis, as well as, in outlining time windows and topographic regions of interest (TOI/ROI) for Experiment 2.

The aim of Experiment 1 was to identify neural correlates of grammatical and ungrammatical pauses in PWDS. Pauses are prosodic breaks that may assist or disrupt syntactic processing. Grammatical pauses are prosodically expected events that facilitate building sentence syntactic structure. In contrast, ungrammatical pauses are prosodically unexpected and may interfere with syntactic processing. Processing the two types of pauses relies on an ability to predict an emerging prosodic pattern and recognise unexpected disruptions. Following the experiment of Besson et al. (1997), where pauses inserted into familiar proverbs (unexpected) evoked larger early negativities in pause-locked event-related potentials than pauses in unfamiliar sentences (less unexpected), it could be inferred that unexpected events may be associated with a larger early negativity compared to expected ones. Based on Besson et al. (1997) it was hypothesised that *unexpected ungrammatical pauses would evoke a larger early negative response compared to expected grammatical pauses.*

Grammatical pauses correspond to the beginning of the closure positive shift (CPS), a neural correlate of prosodic boundaries in speech (Männel & Friederici, 2011; Steinhauer, 2003;

Steinhauer et al., 1999). As the CPS is known to start prior to the onset of the grammatical pause and continue throughout the pause (Kerkhofs, Vonk, Schriefers, & Chwilla, 2007), one could assume that ERP responses to grammatical pauses, as part of the CPS, should be more positive than responses to ungrammatical pauses. Based on this information, it was hypothesised that *ERP responses to grammatical pauses would be significantly more positive than ungrammatical ones around the pause onset time.*

2.2 Method

2.2.1 Participants

Forty adult participants (24 females; mean age: 21.87 years; SD: 5.70) were recruited from the Macquarie University online participant pool. 11 were paid volunteers, the rest participated for course credit. All were native speakers of English. Participants reported normal hearing and an absence of neurological disorders. They were not on medication and had no history of brain injury. The study was approved by Macquarie Ethics Committee.

2.2.2 Materials

Speech stimuli consisted of 103 target sentences that were used in both grammatical and ungrammatical pause conditions and 113 filler sentences. The sentences were semi-automatically extracted from four books by Jane Austen (“Pride and Prejudice”, “Sense and Sensibility”, “Emma”, and “Northanger Abbey”) using Vim text editor (www.vim.org). Sentences were selected based on their syntactic structure. The 103 target sentences were complex sentences where subordinate clauses preceded main clauses. This structure is known to yield longest naturally occurring pauses in speech. The 113 filler sentences had a mix of simple, compound and complex syntactic structures. To prevent habituation to the subordinate + main clause structure, main clauses always preceded subordinate ones in

complex filler sentences. Long sentences were shortened to 12 words per sentence on average. All character names were substituted with other traditional English names to prevent participants from identifying the source texts and focusing attention on specific words. The sentences were then read by an adult male speaker of Australian English and recorded at a sampling frequency of 44100 Hz.

Grammatical pauses

103 complex sentences were used in the grammatical pause condition. Naturally occurring pauses at clause junctures were manually removed and, in their place, 400 ms pauses were automatically inserted using MATLAB 2017a (The Mathworks, Natick, USA). An example of a sentence from the grammatical pause condition is given below:

If we set the example <400 ms pause> many will follow it.

Ungrammatical pauses

The same 103 complex sentences were used in the ungrammatical pause condition. In 50 sentences, 400 ms ungrammatical pauses were inserted into the subordinate clauses, in the rest of the sentences they were inserted into the main clauses. Ungrammatical pauses were functionally defined as pauses that occur in environments requiring continuation of a sentence. The best example of an ungrammatical pause is a pause found between a function and a content word. Function words are words that do not have lexical meanings or have ambiguous lexical meanings. Articles, prepositions, conjunctions, possessive pronouns, relative pronouns, and auxiliary verbs are classified as function words. In the sentences where there were no function-content word environments, ungrammatical pauses were inserted between verbs and predicative adjectives or between high transitively biased verbs and their objects. All in all, there were five transitive verb-object, seven verb-predicative adjective, and

91 function-content word environments. Below is an example of a sentence from the ungrammatical pause condition:

If you do not <400 ms pause> call early, they will be worried.

2.2.3 Experimental procedure

The experiment was conducted in an electromagnetically-shielded sound-attenuated dimly lit room. Participants were seated in a comfortable chair 1.5 m away from the computer screen. The auditory stimuli were presented binaurally via EEG compatible headphones (Etymotics ER2). Presentation software (Version 18.1, Neurobehavioral Systems, Inc., Berkeley, CA) was used for stimulus delivery. Target sentences and filler sentences were presented in a random order separated by 800 ms inter-stimulus intervals. The total duration of the audio presentation was 35 min. A passive listening paradigm was adopted for the study, i.e. participants were not required to perform any tasks while listening to the audio. Before the start of the experiment, participants were informed that they would listen to recorded speech and watch a video clip. No other information was provided. Participants were unaware of the purpose of the experiment.

2.2.4 EEG acquisition

64-channel EEGs were recorded with a sampling rate of 2048 Hz with a BioSemi Active Two system (BioSemi B.V. Amsterdam, Netherlands) using pin-type Active electrodes mounted in a headcap. EEG electrode placement conformed to the international 10/20 standard. Common Mode Sense (CMS) active electrode and a Driven Right Leg (DRL) passive electrode were used as ground electrodes. Electrical activity was recorded from the mastoids via an additional 2 external electrodes and used for re-referencing during data processing. Electrode offsets were kept below 20 μ V. EEG signal acquisition was performed with the ActiView

acquisition program (BioSemi B.V. Amsterdam, Netherlands). Onsets and offsets of pauses, as well as the beginning and the end of each sentence, were marked with timestamps. During EEG acquisition, timestamp information was passed onto the ActiView program via parallel port where they were recorded into the EEG data file.

EEG processing and analysis were performed using the FieldTrip (Oostenveld, Fries, Maris, & Schoffelen, 2011) toolbox for MATLAB (MATLAB version 2017a). EEG data were referenced to the average of the left and right mastoids. Highpass and lowpass filters were applied at 1Hz and 30Hz respectively. The data was then downsampled to 128Hz and cleaned using an automated artefact rejection method (Castellanos & Makarov, 2006). After that, the data was divided into short segments (epochs). Epochs were time-locked to pause onsets, with each epoch starting 500 ms prior to and ending 1000 ms after the pause onset.

2.2.5 EEG data analysis

For the data analysis, cluster-based permutation dependent samples t-test (Maris & Oostenveld, 2007) was performed on the ERPs using pause type (grammatical vs. ungrammatical) as an independent variable. A cluster-based permutation test was selected because it is the most effective way to overcome a multiple comparisons problem in EEG data analysis. Additionally, the permutation model has an advantage over the standard t-test in that it does not require the assumptions of the normal distribution of the dependent variable and of the equality of variance. The cluster alpha used for grouping data samples (channel-time pairs) into clusters was set to .01. Then the Monte Carlo method was applied to identify significant clusters in the data, with the number of randomizations set to 1000.

2.3 Results

The results of the cluster-based permutation t-test revealed that there were one positive and one negative cluster where ERP responses to grammatical and ungrammatical pauses were significantly different between grammaticality conditions. In the positive cluster, ERPs to ungrammatical pauses were associated with a significantly enhanced positivity ($p = .002$) that started 102 ms prior to and ended 94 ms after the pause onset. The distribution of the difference was left-lateralised (see Figure 1).

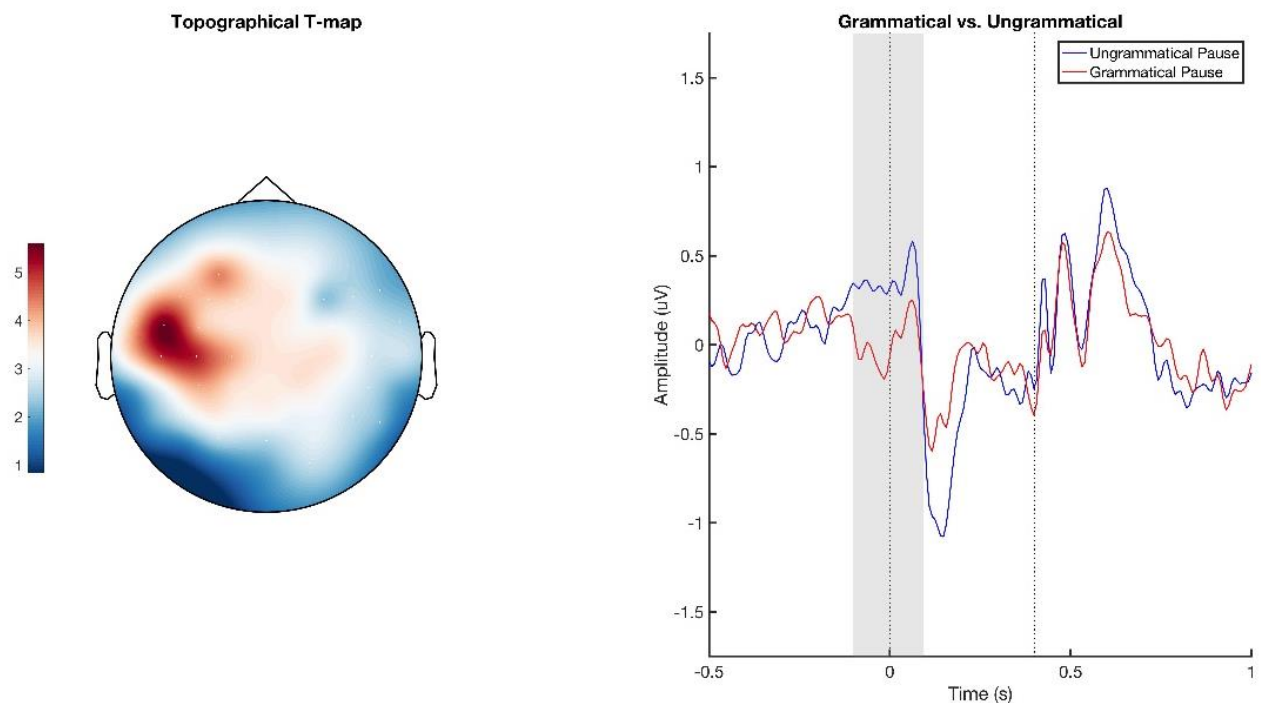


Figure 1. A topographic plot of t-values for the positive cluster (left) and grand averaged ERPs (right) to grammatical and ungrammatical pauses timelocked to pause onset (set at 0). T-values are averaged across the duration of the significant cluster. Colourbar references t-values. ERPs are averaged across all electrodes that are incorporated into the statistically significant cluster at the time of the peak t-value. These electrodes are represented by white markers in the topographic plot. The area between the two hashed lines in the right panel represents the temporal location of the 400 ms pause. The grey-shaded area represents an early (-102-94 ms) time interval where the positive cluster was significant.

In the negative cluster, ungrammatical pauses evoked a significantly larger N1 response ($p = .002$) between 94 and 227 ms after the pause onset. The response was centrally distributed (see Figure 2).

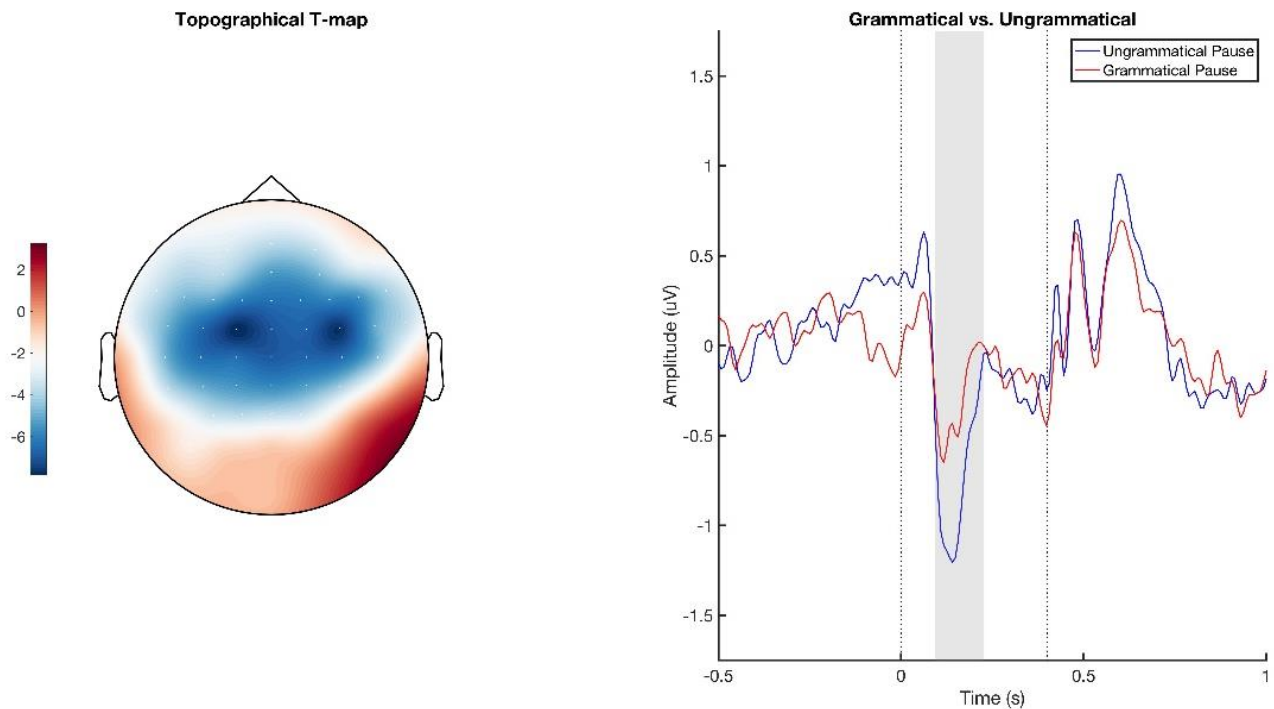


Figure 2. A topographic plot of t-values for the negative cluster (left) and grand averaged ERPs (right) to grammatical and ungrammatical pauses timelocked to pause onset (set at 0). T-values are averaged across the duration of the significant cluster. Colourbar references t-values. ERPs are averaged across all electrodes that are incorporated into the statistically significant cluster at the time of the minimum t-value. These electrodes are represented by white markers in the topographic plot. The area between the two hashed lines in the right panel represents the temporal location of the 400 ms pause. The grey-shaded area represents a late (94-227 ms) time interval where the negative cluster was significant.

2.4 Discussion

The aim of Experiment 1 was to investigate neural responses to grammatical and ungrammatical pauses in a non-stuttering population. The results revealed two time windows where the difference between grammatical and ungrammatical pauses was significant. More precisely, ungrammatical pauses were associated with a significantly higher positivity in the

time window that started approximately 100 ms before and ended around 100 ms after the pause onset (exact time values: -102-94 ms). In addition, ungrammatical pauses evoked a significantly larger N1 response approximately between 100 and 230 ms after the pause onset (exact time values: 94-227 ms).

-100-100 ms time interval

Comparison of ERPs associated with processing of grammatical and ungrammatical pauses revealed that ungrammatical pauses were associated with a significantly stronger positivity with a fronto-central distribution that started approximately 100 ms prior to and ended around 100 ms after the pause onset. A visual examination of the plots revealed that the positivity consisted of two different components: the early component that lasted up to the pause onset and P1 that peaked around 55 ms after the pause onset. While the P1 response to the onset of grammatical and ungrammatical pauses was expected as P1 indexes change in the auditory stimuli (Cone-Wesson & Wunderlich, 2003; Hari & Puce, 2017), the early component where ungrammatical pauses showed higher positivity than grammatical pauses was surprising. In the current study, grammatical pauses were located at syntactic boundaries, a condition that is known to elicit the CPS in listeners (Steinhauer, 2003; Steinhauer et al., 1999), yet, ungrammatical pauses located within syntactic units were associated with a more positive amplitude. This finding cannot be easily explained. The CPS is a large positive waveform that marks sentence prosodic boundaries which often coincide with syntactic boundaries (Bögels, Schriefers, Vonk, & Chwilla, 2011; Steinhauer et al., 1999). Apart from pauses, the CPS is sensitive to other markers of prosodic boundaries, such as lengthening of the preboundary syllable, and pitch change (Männel et al., 2013). In the present study no such markers appeared before the ungrammatical pauses since the ungrammatical pauses were artificially inserted into sentences after the recording.

A potential explanation to the significantly less positive grammatical pauses could be in the known variability of the CPS' onset time, as well as the location of ungrammatical pauses on the waveform. Commonly, the CPS is reported to start at the onset of a pause marking a prosodic boundary or slightly earlier (Bögels et al., 2011; Peter, McArthur, & Crain, 2014; Steinhauer, 2003). However, Kerkhofs, Vonk, Schriefers, and Chwilla (2007) described a CPS that occurred at a later time, between approximately 400 and 800 ms after the offset of the word preceding a pause. Additionally, Pauker, Itzhak, Baum, and Steinhauer (2011) reported a broadly distributed negativity that directly preceded the CPS and peaked at around 50 ms prior to the offset of the verb preceding a pause. The authors interpreted this negativity as a response to the prosodic boundary markers that occurred prior to pauses (pitch change and preboundary lengthening). Thus, if grammatical pauses occurred immediately after the start of the negativity, they may have automatically corresponded to the lowest position on the waveform. In that case, ungrammatical pauses located in the mid-clause positions may have appeared significantly more positive.

Another interesting finding worth mentioning here (albeit it exceeds the -100-100 ms time window) is an apparent absence of the CPS in the grammatical pause condition. According to Steinhauer (2003), the typical duration of the CPS is about 500 ms, with its offset being triggered by the onset of the word after the pause (Bögels et al., 2011; Pauker et al., 2011). Assuming that the CPS in the present study started at around the pause onset, it should have continued through the 400 ms pause till the onset of the next phrase. However, this was not true as the CPS was replaced by a large P1-N1-P2 response to the pause onset. One possibility is that the response to prosodic boundaries can be mediated by pause length. If the pause length is below a certain threshold, it does not affect the CPS production; when the pause length exceeds the above-mentioned threshold, the pause is perceived as a separate auditory event and causes re-analysis of the underlying syntactic structure. Some CPS studies

provide information on pause length that can be correlated with presence or absence of the CPS. For example, Steinhauer (2003) reported a CPS in adults with a pause length of 155 ms. Holzgrefe et al. (2013) reported no CPS in the early boundary condition where the pause length was 297 ms and a CPS in the late boundary condition with 268 ms pauses. Kerkhofs et al. (2007) found a late CPS with 371 ms pauses, Männel and Friederici (2011) reported a CPS in 3- and 6-year-old children with 440 ms pauses, and Glushko, Steinhauer, DePriest, and Koelsch (2016) identified a CPS in non-musicians and no CPS in musicians with 550-600 ms pauses. While, apart from pause length, other factors (e.g. presence of other prosodic boundary markers or participants' age) may contribute to the presence or absence of the CPS, it appears there is a tendency, at least in some adults, for a reduced CPS if the pause is comparatively long. Future research may aim to identify the pause length threshold after which the CPS is disrupted.

This study was the first to report that ungrammatical pauses within clauses are associated with significantly larger positivity than grammatical pauses. As grammatical pauses occur immediately after the negativity that precedes the CPS and may be associated with prosodic boundary markers, they may appear less positive compared to ungrammatical pauses found in mid-clause positions. A supplementary finding is that the CPS does not develop fully as it may be interrupted by a relatively long pause, however, more research of this phenomenon is needed.

100-230 ms time interval

In the late time window, ungrammatical pauses evoked a significantly larger negative centrally distributed response that peaked at 140 ms after the pause onset. A negative response to an auditory stimulus change in that time window is referred to as an N1. As it indexes auditory change, the N1 is evoked equally by sound onset and sound offset. The N1

is known to characterise stimulus intensity and timing (Näätänen & Picton, 1987). For example, Palmer and Musiek, (2013) found that the amplitude of the N1-P2 response to a gap in noise was mediated by the gap duration, i.e. longer durations corresponded to larger amplitudes. Similarly, Pratt, Starr, Michalewski, Bleich, and Mittelman (2007) reported that the intensity level of the pre-gap noise affected latencies and amplitudes of evoked responses time-locked onto gap onsets. This, however, could not explain the present findings as pause durations were the same for both conditions. Similarly, the volume of the recorded speech was normalised, which, at least partly, controlled for pre-pause sound intensity variability between the two conditions.

The only difference between the two conditions in the present study was in the level of expectancy between the two types of pauses. Based on their knowledge of prosodic patterns, participants were likely to anticipate grammatical pauses at syntactic junctures, while ungrammatical pauses were completely unexpected. It is possible that differences in the N1 amplitude between the two conditions could be mediated by the level of stimulus expectancy, with unexpected pauses eliciting larger ERPs. This goes in line with the theory of predictive processing in language comprehension. The theory posits that the brain continuously creates predictions about upcoming stimuli which can affect how those stimuli are processed (Bar, 2007). This combination of bottom-up and top-down processing accounts for a fast speech processing speed that cannot be explained if only bottom-up processes are considered. Specifically, Dikker and Pylkkänen (2011) examined whether the brain generated word-form estimates based on presented pictures and how they affected subsequent word processing. They reported that the M100 response (a MEG equivalent of N100) to words that violated lexical-semantic expectations was significantly larger compared to the words that were congruent with participants' expectations. Similarly, Dikker, Rabagliati, Farmer, and Pylkkänen (2010) analysed how prediction of a word's syntactic category affected processing

of a word that violated that prediction. They found an enhanced M100 to violating predictions words whose form was typical of their syntactic category (e.g. the word ‘princess’ in “The beautifully *princess...”). Overall, these findings support the theory that expectancy plays an important role in language processing and accounts for rapid processing of prosodic, syntactic, and semantic violations.

Predictive processing helps to explain the results of another study that came closest to examining ungrammatical pauses. (Besson et al., 1997) studied ERPs to pauses inserted before two sentence final words. They used two types of stimuli: well-known French proverbs and unfamiliar sentences. The ERP response to pauses included a large negative component that peaked around 200 ms and a large positive component that peaked at around 350-400 ms after the pause onset. The amplitude of the ERP response was larger for familiar than unfamiliar sentences. This can possibly be explained by the fact that participants formed stronger expectations about upcoming words in well-known proverbs which lead to enhanced responses when those expectations were violated by an inserted pause.

In the current study, participants were much more likely to expect grammatical pauses than ungrammatical ones due to their extensive experience with the language. As was discussed earlier, grammatical pauses occur at prosodic boundaries and are accompanied by other boundary markers, such as final syllable lengthening and pitch change. Grammatical pauses are processed by the brain even when they are not actively attended to as shown by Peter's et al. (2014) CPS study. Ungrammatical pauses, on the other hand, cannot be predicted from the context as no prosody or pitch changes precede them. As a result of their high ‘unexpectedness’, ungrammatical pauses elicited larger N1 responses compared to grammatical pauses. The results of the present study show that the auditory N1 response reflects not only physical properties of speech stimuli (which were kept the same – a 400 ms absence of speech) but also listeners’ predictions about those stimuli.

Perhaps, it is worth mentioning here that some prosodic violation studies have reported positive, as opposed to negative, responses. For example, Paulmann, Jessen, and Kotz (2012) examined linguistic, emotional and emotional-linguistic prosodic violations. They found that while linguistic and emotional violations evoked late positivities, an emotionally-linguistic violation evoked an early positivity with a latency of 170 ms after the violation onset. Similarly, Zioga, Di Bernardi Luft, and Bhattacharya (2016) reported a relatively early positive response with a latency of 150 ms to prosodic expectancy violation in question-statement pairs. One explanation of the polarity difference could be that it is determined by the type of an auditory input. While the abovementioned studies used intonation changes, in the present study used pauses that constitute a much larger acoustic change. The encouraging finding is that Besson et al. (1997) who similarly used pauses, also reported negative responses.

One possible limitation of this study is that no control for attention was implemented. Since participants were instructed to watch a silent movie and listen to the speech, it was not clear how exactly their attention was distributed between the two tasks. Some may have focused on the movie, while others may have attended to speech. Since attention is known to modulate the N1 response (Martin, Tremblay, & Korczak, 2008), it would be interesting to examine whether controlling for attention would in any way change the results.

In this study, grammaticality was approached as an all-or-nothing phenomenon which allowed us to create the two opposite conditions: highly predictable grammatical pauses marking prosodic boundaries and highly unpredictable ungrammatical pauses located predominantly between function and content words. Another way to study grammaticality is as a continuum. For example, if we assume that pauses between noun and verb phrases are also unexpected and, to a certain extent, ungrammatical, we can possibly measure the degree

of ‘unexpectedness’ required to create a significant effect. This approach can be implemented in the future research.

To sum, the results revealed that ungrammatical pauses produce a significantly larger N1 response compared to grammatical ones, which shows that grammatical and ungrammatical pauses are rapidly discriminated between during speech perception. These findings can best be explained from the perspective of the predictive processing theory which accounts best for their fast processing speed. Since ungrammatical pauses act as prosodic violations, N1 can be viewed as a neural marker of prosodic processing. This assumption will be used in formulation of the hypothesis for Experiment 2.

Chapter 3: Neural correlates of prosodic perception in PWS and PWDS (Experiment 2)

3.1 Hypothesis

The aim of Experiment 2 was to investigate whether adults who stutter (AWS) process speech prosody differently from adults who do not stutter (AWDS). Prosody comprises speech rhythm and intonation. As PWS experience difficulties with general rhythm perception (Chang et al., 2016; Wieland et al., 2015), they are likely to have problems with perceiving speech rhythm, which is crucial for prosodic processing. Hence, it is possible that prosodic processing in PWS may be impaired.

The results of Experiment 1 revealed that prosodically unpredictable ungrammatical pauses evoked larger N1 amplitudes compared to predictable grammatical ones. The results suggest that N1 acts as a neural marker of pause grammaticality and is reflective of an individual’s predictive processing ability. This marker could be used to test perception of prosodic

violations in a stuttering population. If AWS, indeed, process prosody differently from AWDS, the neural response to prosodic violations would be expected to differ between the two groups. Based on the results of Experiment 1, it was hypothesised that *AWS would show a smaller N1 amplitude to unexpected ungrammatical pauses due to their impaired predictive processing of speech prosody*. As grammatical pauses are highly predictable and facilitate syntactic structure analysis, it is unlikely that AWS would experience difficulties in processing them. *Thus, it was hypothesised that the N1 response to expected grammatical pauses would be similar across the two groups.*

Another finding of Experiment 1 was that the neural activity preceding ungrammatical pauses and immediately following them, was significantly more positive than that for grammatical pauses. Since the early positivity was determined by slow potentials that developed in response to a suprasegmental prosodic structure (e.g. the CPS), AWS would unlikely differ from AWDS in that time interval as AWS would have enough time to compensate for possible early speech processing deficits. Thus, it was hypothesised that *no difference between AWS and AWDS in the -100-100 ms time interval would be found.*

3.2 Method

3.2.1 Participants

Participants were 15 AWS (5 females; mean age: 38 years; SD: 16 years) and 15 age- and sex-matched AWDS (Mean age: 36 years, SD: 17 years). There was no significant age difference between the two groups ($t(28)=.29$, $p=.78$). Participants did not have any neurological disorders other than stuttering, were not on medication and reported no history of brain injury. Participants underwent a hearing screening test at 500 Hz, 1000 Hz, 2000 Hz, 3000 Hz. 28 participants demonstrated normal hearing, while two participants experienced a mild hearing loss in the 3000 Hz range. However, since a typical male voice fundamental

frequency falls below 3000 Hz, a mild hearing loss in that range should not have affected speech comprehension during the experiment (Hollien & Shipp, 1972). All participants were right-handed. Participants received monetary compensation for their time. The study was approved by Macquarie Ethics Committee.

Table 1. AWS background information.

ID	Gender	Age	Average stuttering severity (self-rated)	Therapy	Relatives who stutter	OASES overall impact score
Subject 1	M	43	3	Yes	No	Moderate/Severe
Subject 2	F	26	3	Yes	Yes	Moderate
Subject 3	F	68	0 ¹	Yes	No	Mild
Subject 4	M	27	4	Yes	Yes	Moderate
Subject 5	F	22	5	Yes	Yes	Moderate
Subject 6	F	66	4	Yes	Yes	Moderate/Severe
Subject 7	F	24	3	Yes	Yes	Moderate
Subject 8	M	33	7	Yes	Yes	Moderate/Severe
Subject 9	M	48	4	Yes	Yes	Mild/Moderate
Subject 10	M	40	2	Yes	No	Moderate
Subject 11	M	33	5	Yes	No	Moderate
Subject 12	M	64	2	Yes	No	N/A ²
Subject 13	M	26	4	Yes	Yes	Mild/Moderate
Subject 14	M	26	3	Yes	No	N/A ³
Subject 15	M	22	3	Yes	Yes	Moderate

¹ – Subject reported using a speech technique that controlled her stuttering.

² – Subject did not answer the required minimum of questions for a general score could be calculated.

³ – Subject chose not to fill out the questionnaire.

3.2.2 Stimuli, Experimental Procedure and EEG acquisition

The stimuli, the experimental procedure, and the EEG acquisition were as per Experiment 1.

3.2.3 EEG analysis

The topography and latency of significant responses from Experiment 1 was utilised to define time (TOI) and regions of interest (ROIs) for Experiment 2. Experiment 1 identified two time windows (-100-100 ms and 100-230 ms) where statistically significant effects of grammaticality were found in the ERP analysis. These were used as TOIs for Experiment 2. ROIs were defined as electrodes where response differences between ungrammatical and grammatical pauses within the specified TOI were largest (above the 75th percentile). This was done because both positive and negative responses had a widely distributed topography and since Experiment 2 had fewer participants, comparisons between the groups would likely be insensitive to true differences if correction across the whole search space and time was conducted. As a result, the following electrodes were selected as ROIs for the early time window data analysis: Fp1, AF3, F1, F3, FC1, C1, C3, C5, CP1, P3, CPz, FC4, C4, C6, CP4, P4. A, while the following electrodes were selected as ROIs for the late time window analysis: F1, F3, FC3, FC1, C1, AFz, Fz, F2, F4, FC6, FC4, FC2, FCz, Cz, C2, C4. A 2x2 mixed-design analysis of variance (ANOVA) was then performed using fluency (AWS vs. AWDS) as a between-groups factor and a pause type (grammatical vs. ungrammatical) as a within-groups factor. The mean ERP amplitude within the a priori defined ROI/TOI was the dependent variable.

3.3 Results

The results showed that there was a significant fluency x pause type interaction ($F(1, 28) = 4.48, p = .04$) in the 100-230 ms time window. As it can be seen from Figure 3, AWDS showed a much higher negative amplitude for ungrammatical pauses than AWS, while the response to grammatical pauses was very similar for both groups. There was a significant

effect of a pause type ($F(1, 28) = 22.42, p < .01$), while the main effect of fluency was not statistically significant ($F(1, 28) = 1.95, p = 0.17$).

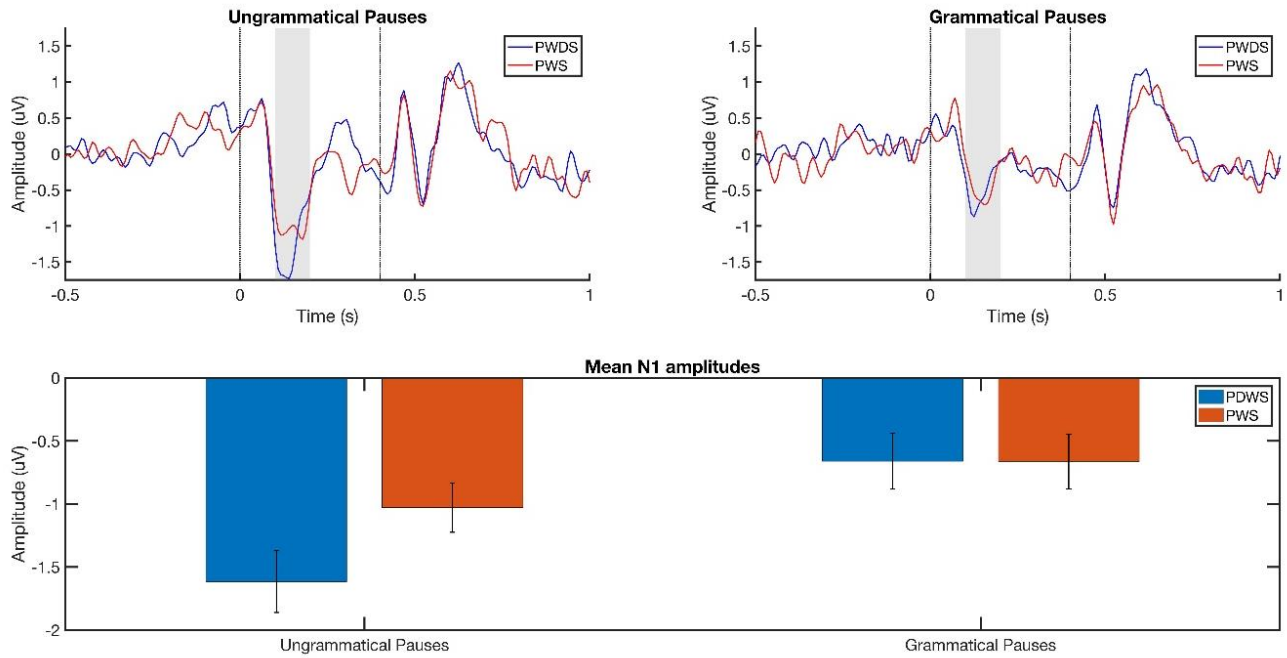


Figure 3. Top row: Grand-averaged ERPs (averaged across electrodes in the ROI) to ungrammatical pauses in PWS and PWDS (left) and grammatical pauses in PWS and PWDS (right) timelocked to pause onset (set at 0). Bottom row: Bar charts showing mean N1 amplitude in response to ungrammatical pauses (left) and grammatical pauses (right) in PWS and PWDS.

In the -100-100 ms time window, the pause type main effect ($F(1,28) = 3.76, p = 0.06$), the fluency main effect ($F(1,28) = 0.51, p = 0.48$) and the fluency x pause type interaction ($F(1,28) = 0.58, p = 0.45$) were not significant.

3.4 Discussion

This study examined neural responses to grammatical and ungrammatical pauses in AWS and AWDS. Using the time windows where significant differences between grammatical and ungrammatical pauses were found in Experiment 1 as a priori constraints to the analysis, we compared neural responses to both types of pauses in AWS and AWDS in -100-100 ms and 100-230 ms time windows relative to pause onset. As expected, no differences between the

two groups were found in the early time window, which is associated with processing of the pre-pausal speech as well as the early response to the pause onset. The results for analysis of the 100-230 ms time window showed that AWS had a significantly reduced N1 amplitude in response to ungrammatical pauses compared to AWDS. There was no difference between the groups in the response to grammatical pauses. As expected, AWS did not differ from AWDS in processing expected grammatical pauses, but they were different from AWDS in how they processed prosodic violations represented by ungrammatical pauses.

As was established in Experiment 1, ungrammatical pauses elicited an enhanced N1 response compared to grammatical pauses in a non-stuttering population. This finding is interpreted from the perspective of predictive processing which suggests that individuals incrementally form expectancies about upcoming stimuli, which subsequently affect processing of those stimuli when they occur in speech (Bar, 2007; Dikker & Pylkkanen, 2011; Dikker et al., 2010; Pickering & Garrod, 2013). Similarly to Experiment 1, the results of Experiment 2 showed that both AWS and AWDS had an enhanced N1 response to ungrammatical pauses. However, the amplitude of the N1 response to ungrammatical pauses was significantly reduced in AWS. The amplitude of the N1 response to grammatical pauses was similar across both groups. This indicates that AWS may be worse at discriminating between grammatical and ungrammatical pauses compared to AWDS. From the predictive processing point of view, the results support the hypothesis that AWS may have a reduced ability to predict upcoming prosodic elements, which may affect their ability to respond to them when they are encountered in speech.

As far as I am aware there are no studies that have examined pause perception in a stuttering population. In fact, there are very few studies that have investigated prediction violations in speech perception in PWS. Of those that have, most report differences between stuttering and non-stuttering populations predominantly in the late (cognitive) ERP components (Morgan,

Cranford, & Burk, 1997; Usler & Weber-Fox, 2015; Weber-Fox, Hampton Wray, & Arnold, 2013; Weber-Fox, Spruill, Spencer, & Smith, 2008). To my knowledge, only one study that examined neural responses to auditorily presented sentences, reported differences in early ERP components between stuttering and non-stuttering populations. Specifically, Weber-Fox et al., (2013) found an increased N1 amplitude to syntactic (phrase structure) violations in CWS compared to CWDS. Our findings showed a reverse effect, with AWS producing lower N1 amplitudes in response to prosodic (speech rhythm) violations manifested as ungrammatical pauses. This discrepancy is intriguing. One possible explanation for this difference is that that CWS may be more sensitive to syntactic violations and less sensitive to prosodic ones compared to CWDS. This conclusion falls in line with the theory of PWS' internal timing deficits that may affect PWS' speech rhythm perception and result in poorer predictive processing of speech prosody. Deficits in rhythm perception, on the other hand, are less likely to affect syntactic processing, hence, PWS may respond to syntactic violations similarly to PWDS. Still, there is no good explanation as to why N1 was enhanced to syntactic violations in Weber-Fox' et al. study. Perhaps, other unknown factors contributed to the outcome of their study.

Other studies that investigated syntactic violations in PWS, have found no differences in the early ERP components between stuttering and non-stuttering groups (Usler & Weber-Fox, 2015; Weber-Fox & Hampton, 2008). The results of these studies suggest that PWS may be similar to PWDS in the early phases of syntactic processing. This is an important finding as it confirms (considering that semantic processing occurs later) that N1 amplitude differences between grammatical and ungrammatical pauses in the current study are likely to arise from prosodic violations as opposed to syntactic processing.

An interesting, albeit a speculative, perspective on the cause of PWS' prosody perception deficits comes from ERP studies of brain-stem auditory evoked potentials (BAEPs). The

results of those studies suggest that AWS are poorer at making predictions about speech due to impaired low-level auditory processing. Specifically, there are two studies that have found systematic differences between AWS and AWDS in BAEPs. Blood and Blood (1984) reported that, in response to clicks, latencies of Wave III and Wave V were significantly longer in AWS than in AWDS. Also, interpeak latencies were abnormally prolonged in 62% stuttering participants. Another study by Tahaei, Ashayeri, Pourbakht, and Kamali (2014) did not find differences in BAEPs produced in response to clicks, however, they reported that speech-related BAEP latencies were increased in AWS. The authors attributed this finding to timing disturbance in the auditory pathways in AWS. Hypothetically, these tiny abnormalities at a low-level of auditory processing could affect higher-order auditory processing, particularly, in the case of complex stimuli, such as speech. The question is then if AWS have low-level auditory processing deficits, why AWS perceived grammatical pauses similarly to AWDS. It is possible that AWS develop compensatory strategies that work when the auditory input is predictable, as it was found with processing of grammatical pauses. However, those strategies may break down when the auditory input is unpredictable, as it was with ungrammatical pauses. This explanation is very speculative though as there is lack of research in the field of auditory brain-stem responses and stuttering.

Indirect support for the idea that prosodic processing may be impaired in AWS due to underlying internal timing deficits comes from studies of brain functional connectivity during speech perception tasks in PWS. Specifically, Halag-Milo et al. (2016) performed an fMRI scan while participants were listening to short poems. Their results showed that AWS had a stronger activation in the right inferior frontal gyrus (IFG) and left Heschl's gyrus compared to control subjects. As the right IFG is commonly activated during speech production in PWS, the authors concluded that a single imbalanced network may have caused both speech perception and production deficits in PWS. Another fMRI study may have identified the

neural mechanism that explains anomalous functioning of the left Heschl's gyrus in PWS. Lu et al. (2016) found that PWS had a weaker resting-state functional connectivity between the left Heschl's gyrus and the left inferior cortex/anterior insula involved in speech production, while the strength of this connection correlated with speech perception performance. The authors concluded that anomalous functioning of the speech motor area affected performance of the temporal auditory area which lead to speech perception deficits. The results of both of these fMRI studies fit with the motor theory of speech perception that posits that speech perception involves perception of the speaker's vocal tract gestures (as opposed to phonology and prosody). The theory stresses that speech production and perception are closely related due to recruitment of the same production network during speech perception (Galantucci et al., 2006; Liberman & Mattingly, 1985). In accordance with this theory, AWS's deficits in speech perception are caused by dysfunctions at a speech production level. This implies that timing deficits found in speech production in PWS may also extend to speech perception.

Due to impaired predictive processing, PWS may be less efficient in automatic first-pass parsing of speech input. In the current study, decreased parsing efficiency manifested in a reduced N1 amplitude in response to unpredictable ungrammatical pauses. One of the consequences of decreased parsing efficiency could be that PWS require more time to successfully process speech input. The idea that PWS may need more time to process speech is supported by delayed auditory feedback studies. Research has shown that stuttering is significantly alleviated if PWS hear their own speech with a short delay. Delays that are known to be beneficial for PWS' speech range between 13-200 ms, with most common being about 50-80 ms (Lincoln et al., 2006; Van Borsel, Sierens, & Pereira, 2007). If auditory processing is slower in PWS, it is possible that this delay is required for them to "catch up" with processing their own or someone else's speech. In the current study, no statistical

analysis was performed on latency data, however visual inspection suggested that the N1 peak was slightly delayed in PWS.

It is also possible that due to less efficient speech parsing, PWS required more attentional resources to process speech input than the passive paradigm with a distractor task (used in the current study) could afford them. In other words, as part of PWS' attentional resource was engaged with the movie, the remaining free part was insufficient for effective speech input processing, which was not the case for PWDS. This theory is supported by the fact that auditory N1 is known to be attention mediated, with higher amplitudes corresponding to higher attention engagement (Näätänen, Risto & Picton, Terence, 1987). Thus, if AWS were slower to process speech than AWDS, it could take them longer to perceive prosodic violations in the form of ungrammatical pauses and attend to them, which could lead to a lower N1 amplitude.

There have been no studies of attentional demand in speech perception in PWS. However, the results of production studies suggest that this is a possibility. Maxfield et al. (2016), using a dual-task paradigm, investigated whether speech production drew a disproportionate amount of resource away from a secondary task in PWS. Specifically, in their study, participants named pictures in the presence of phonological, semantic and unrelated distractors and, at the same time, performed an oddball task while ERP responses to the target tones (with early or late onsets) were recorded. The authors were particularly interested in the P3 response that indexes availability of attentional resources. They found that while PWDS' P3 responses were present for all conditions, PWS' P3 responses were significantly attenuated or absent for half of the conditions. Maxfield et al. (2016) concluded that resolving language ambiguities required a significantly larger amount of attentional resources for PWS compared to PWDS. If speech production in PWS requires extra attentional resources, it is plausible that speech perception does so too, which could help to explain the results of the present study.

One of the possible limitation of this study was a very wide age range of participants, with the youngest and the oldest participants being 22 and 68 years old respectively. Even in the absence of hearing impairment, older adults are known to process auditory input differently. Due to loss of cochlear neurons and neurons in the auditory centres of the brain, older adults experience a disproportionate loss of the ability to process speech as opposed to non-speech sounds (Howarth & Shone, 2006). According to the inhibitory deficit hypothesis, older adults have a diminished ability to inhibit irrelevant auditory information which results in changed amplitudes and latencies of auditory ERPs (Stothart & Kazanina, 2016). There is a surprising lack of data on the progress of stuttering disorder into older age. Our own data, based on AWS' self-reports, suggests that stuttering severity may diminish with age, perhaps, because older AWS develop better coping strategies and use fluency-enhancing techniques that suit them best. It is unknown, however, if there are any changes on a neurological level associated with aging in PWS. While having participants of a broad age range may not have affected the results of this study due to the fact that there was an age-matched control group, to ensure a better understanding of age-related changes in stuttering, studying PWS groups that are less heterogeneous in age demographics would be advisable. Future studies could consider looking into neurology of stuttering in older people as, to date, there has been no such research.

This study focused on examining only early ERP components of pause perception as we had clearly defined epochs of interest determined by the results of Experiment 1. While looking into early sensory responses is very beneficial for pinpointing physiological differences between stuttering and non-stuttering populations, examining later cognitive ERPs would reveal how differences at an early processing stage could contribute to subsequent re-evaluation of the sensory information. Intriguing findings were presented by Friederici, Cramon, Yves, and Kotz (1999) who examined ERPs to semantic and syntactic violations in

patients either with left frontal cortical lesions or basal ganglia lesions. They found that while patients with basal ganglia lesions did not differ from controls, patients with left frontal cortical lesions showed absence of the early negativity but a normal P600 response to syntactic violations. Grammaticality judgements of sentences with both types of violations were the same across both patient groups and controls. The authors hypothesised that the first-pass parsing, indexed by the early ERP negativity, may not be necessary for subsequent processing. Future research could focus on how differences in early ERPs between PWS and PWDS translate into differences in cognitive ERPs as speech processing continues.

The present study examined neural correlates of PWS's and PWDS' processing of prosodic violations. While it was found that PWS and PWDS process grammatical and ungrammatical pauses differently, it is not known whether PWS actually perceive prosodic violations differently from PWDS. Behavioural studies would be needed to establish that relationship.

In conclusion, this experiment was the first to investigate grammatical and ungrammatical pause perception in stuttering and non-stuttering adults. While both groups did not differ in their responses to grammatical pauses, stuttering adults showed an attenuated early negativity to ungrammatical pauses. This finding was interpreted as a diminished ability to discriminate between prosodic events during early stages of speech processing and attributed to failure to successfully predict prosodic elements in speech. In order to determine the cause of impaired predictive processing in AWS, other studies that investigated speech perception in PWS were examined. Deficits in low-level auditory processing, an overlap of brain areas involved in speech production and speech perception, and delayed auditory processing could potentially be implicated in impaired prosody perception. One implication of impaired predictive processing may be a slower first-pass parsing of speech, which is supported by delayed auditory feedback studies. A possibility of attentional involvement in AWS' altered prosodic processing could not be excluded. Future research may focus on how exactly early processing

deficits affects later stages of speech processing, as well as examining behavioural differences in prosody perception between PWS and PWDS.

Summary

The two parts of this thesis have made important contributions to: 1) identifying neural correlates of pause perception in a non-stuttering population and 2) understanding prosodic processing in PWS. The results of the first part provided physiological data on pause processing in a non-stuttering population that was used in investigation of prosodic violation processing in PWS.

Experiment 1 compared neural processing of grammatical and ungrammatical pauses. To date, very few neurophysiological studies have attempted to examine differences in neural processing related to grammatical and ungrammatical pause perception. The results show that ungrammatical pauses in mid-clause positions are associated with higher positivity than grammatical pauses marking prosodic boundaries. Additionally, this study is first to show that prosodically unexpected ungrammatical pauses evoke larger N1 responses than prosodically expected grammatical pauses. These results suggest that prosodic violations are rapidly differentiated from non-violations in speech processing. The latter finding was employed in the second part of the thesis.

The second part of the thesis has made an important contribution to understanding of PWS' processing of expected and unexpected prosodic events. Firstly, most studies that investigated speech perception in PWS to date have focused either on single word processing or syntactic-semantic violations. There have been no neurophysiological studies that have examined prosodic processing in PWS. In this respect, this study is the first to show that PWS have different neurophysiological responses to prosodic violations to PWDS.

Secondly, those studies that looked into auditory sentence processing in PWS have reported differences between stuttering and non-stuttering populations in the late cognitive components that index re-analysis of syntactic and semantic information. In contrast, the present study identified the N1 response as a neural marker of prosodic violation which suggests that differences between PWS and PWDS in prosodic processing occur early during speech perception and are precognitive in nature. The implication of this finding is that PWS may have difficulties in perceiving meaningful prosodic cues during initial speech parsing that could affect their subsequent syntactic and semantic structure analysis.

We interpret differences between PWS and PWDS from the perspective of impaired predictive processing. As PWS experience difficulties with non-speech rhythm perception, they are likely to have problems with speech rhythm perception which is very complex and requires a superimposition of periodicity. The ability to predict speech rhythmic elements is essential for successful prosodic processing. As a result, prosody perception in PWS may be impaired. This conclusion is very important as it connects PWS' rhythm disturbance at the perception level to the rhythm disturbance at the production level, thus providing support to the theory of a common underlying mechanism that causes deficits in the perception and production domains in developmental stuttering.

The findings of the present study do not solve the mystery of stuttering, yet, they contribute to our understanding of problems that may arise from impaired prosodic processing.

Specifically, the ability to correctly parse speech is extremely important for language acquisition. If CWS have difficulties with prosody perception, their language development may be delayed or they may develop dysrhythmic speech (cluttering). Similarly, the ability to perceive prosodic patterns is essential for foreign language learning. Daily communication may also be affected if PWS have problems differentiating between emotional prosodic patterns. Knowing these problems may stem from impaired prosodic processing, developing

new techniques aimed at explicit training in various types of prosody may be needed. For example, an explicit explanation of sentence prosodic patterns would benefit PWS with concomitant cluttering. Training in recognition of emotional prosody may be beneficial for stuttering children. This may not resolve stuttering per se but may help to improve day-to-day communication of people who stutter.

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