Non-speech behaviours in neurogenic stuttering

Master thesis

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Abstract

Neurogenic stuttering is characterised by the occurrence of speech disfluencies following neurological brain damage. While people with a developmental onset of stuttering typically present with non-speech behaviours associated with their stuttering (e.g. eye blinking, facial grimacing), it has been argued that such behaviours are absent in people with a neurological onset of stuttering (Helm-Estabrooks, 1999). However, a number of case-studies suggested otherwise (e.g., Tani and Sakai (2010), Vanhoutte et al. (2014)). This study aimed to investigate, for the first time, the non-speech behaviours in a larger group of people with neurogenic stuttering.

This study consisted of 22 participants with a diagnosis of neurogenic stuttering and a control group of 17 healthy older adults. Their speech was analysed by annotating all stuttering-like disfluencies (SLD), other disfluencies (OD) and non-speech behaviours (NSB). For each non-speech behaviour, duration and severity was also coded.

The results showed that the frequency of occurrence of non-speech behaviours was higher within the neurogenic stuttering group (M = .12, SD = .13) compared to the control group (M = .02, SD =.19). The duration and severity of the NSBs were also different between the groups. SLDs were a significant predictor of proportion of NSBs ($\beta = .4$, $t = 3.39$, $p < .01$) and duration of NSBs ($\beta = .43$, $t = 2.9$, $p < .01$), as well as of a score combining all three NSB measurements ($\beta = .5$, $t = 4.05$, $p < .001$). In a model without outliers, the severity of NSBs was also significantly predicted by SLD proportion ($\beta = .67$, $t = 5.46$, $p < .001$). Within the neurogenic stuttering group, SLDs were the most important predictor of the combined NSB score ($\beta_{SLD} = .41$, $t_{SLD} = 3.2$, $p_{SLD} < .01$) and the proportion of NSBs ($\beta = .34$, $t = 2.32$, $p = .03$).

The results show that non-speech behaviours do occur more frequently and are more severe in people with neurogenic stuttering compared to a control group of healthy speakers. This is in contrast with previous publications stating that non-speech behaviours do not present in neurogenic stuttering. Time post-onset and emotions and attitudes associate with speech were not significant predictors of the proportion of NSBs, contradicting the theory that NSBs develop as a reaction to stuttering.

1. Introduction

Stuttering is a well-known speech disorder. Because of the high incidence of childhood stuttering (5% or even higher), most people know at least one person who stutters and have a clear image of what stuttering entails (Yairi & Seery, 2015). Although stuttering is a common disorder and a lot of research is being done about this subject, a lot remains uncertain. There are, for example, many theories about the underlying cause of stuttering, but they have yet to be confirmed (Yairi & Seery, 2015). In contrast to developmental stuttering, with an onset in childhood, neurogenic stuttering occurs following neurological disorders, typically in adults. Research about neurogenic stuttering is rare, and as a result even less of this disorder is understood.

This thesis focusses on characteristics of neurogenic stuttering, and the non-speech behaviours (NSBs) of people with neurogenic stuttering in particular. A definition of stuttering and the classification of stuttering disorders will be described in this introduction. Because most of the research about stuttering characteristics and non-speech behaviours of stuttering focusses on developmental stuttering, it is important to look at developmental stuttering to understand neurogenic stuttering. Therefore, an overview of research about developmental stuttering will be given next. Thereafter, research about neurogenic stuttering will be described. Finally, the research questions of this thesis will be listed and motivated.

1.2 Definition and classification of stuttering disorders

There is no consensus about the definition of stuttering (Yairi & Seery, 2015). Stuttering can be defined purely as a speech disorder where speech is characterised by disfluencies. Several problems arise in this definition however. Disfluencies also occur in normally fluent speech and are therefore not exclusive to speech of people who stutter (PWS). When listening carefully to seemingly fluent speech, it becomes apparent that this is often interrupted by disfluencies (Logan, 2015; Yairi & Seery, 2015). Similarly, there is evidence that fluent speech of PWS differs from fluent speech of normally fluent speakers (NFS). The distinction between fluent speech and stuttered speech might therefore be more difficult than just the occurrence of disfluencies (Yairi & Seery, 2015). Another problem with a definition that only includes speech characteristics is that PWS experience more problems associated with stuttering than the disfluencies alone. Thus, a more complex definition of stuttering would seem appropriate, which includes not only the speech characteristics of PWS but also NSBs, for example: “A speech disorder affecting the fluency of production, often characterized by repetitions of sounds and blocking of the articulation of words. Severer forms may be associated with facial grimacing, limb and postural gestures, involuntary grunts, or impaired control of airflow. The severity of symptoms may vary with the speaker’s situation and audience”(Yairi & Seery, 2015, p. 15). This definition implies that associated behaviours only appear in more severe forms of stuttering and doesn’t mention emotional features associated with stuttering. However, the symptoms of PWS vary and not everybody who stutters shows NSBs or emotional problems that are associated with stuttering. It is difficult to include all possible problems associated with stuttering in a definition and still have a definition that describes the stuttering disorder of all people who stutter. Yairi and Seery (2015) propose therefore that it might be best to only include the speech characteristics of stuttering in the definition, because stuttering is diagnosed on the basis of speech characteristics. Thus, the used definition is that of Yairi and Seery (2015, pp. 18, 19): “we define stuttering as articulatory

gestures in a holding pattern (repetition, prolongation, block) in an attempted delivery of syllables (including single-syllable words) or elements of syllables", but included with: next to speech characteristics, emotional features and NSBs associated with stuttering might be present.

Different types of stuttering exist. The type of stuttering which starts at an early age, developmental stuttering, is most well-known. According to the Diagnostic and Statistical Manual of Mental Disorders 5 (DSM-5), a fluency disorder with an onset during or after adolescence is called adult-onset fluency disorder (American Psychiatric Association, 2013). Adult-onset fluency disorders may be caused by neurological insults, medical conditions and mental disorders. However, there are several reported cases of children who began stuttering after a neurological event. They can therefore not be diagnosed with developmental stuttering and adult-onset-stuttering doesn't seem to be the right term (Van Borsel, 2014). Therefore the differentiation of stuttering types suggested by Costa and Kroll (2000) may be more accurate. They differentiate between developmental stuttering, persistent developmental stuttering and acquired stuttering. Developmental stuttering is seen as "stuttering with a gradual onset in childhood as a disturbance in the normal fluency and time patterning of speech" (Costa & Kroll, 2000, p. 1850). If the developmental stuttering does not resolve spontaneously or with speech-therapy, it becomes persistent developmental stuttering. Costa and Kroll (2000) divide acquired stuttering into two types: neurogenic and psychogenic. Ashurst and Wasson (2011) also differentiate between developmental, neurogenic and psychogenic stuttering, as do Prasse and Kikano (2008). Neurogenic stuttering is described as "typically the result of nerve or traumatic brain injury" (Ashurst & Wasson, 2011, p. 576) whereas psychogenic stuttering is described as stuttering with a sudden onset after emotional trauma or stress. Van Borsel described all different terminologies that have been used in the literature of acquired stuttering. He proposed the following terminology (Van Borsel, 2014, p. 46):

"Acquired stuttering: Most general term to refer to fluency problem that is not of developmental origin in an individual with no pre-existing stuttering.

- *Psychogenic stuttering:* Subtype of acquired stuttering; dysfluency, associated with a psychological problem or an emotional trauma. A Psychopathological diagnosis need not be required.
- *Neurogenic stuttering:* Subtype of acquired stuttering; dysfluency associated with acquired brain damage in an individual with no pre-existing stuttering."

In this thesis, the classifications, terminology and definitions proposed by Van Borsel (2014) are used. Van Borsel (2014) distinguishes several other terms such as stuttering associated with acquired neurological disorders (SAAND), thalamic stuttering and drug-induced stuttering. SAAND includes acquired stuttering, but differs because it also includes individuals who have a history of developmental stuttering which is worsened or returned due to the neurological disorders. Thalamic stuttering can be seen as a subtype of neurogenic stuttering, resulting from damage in the thalamus. Since it is a subtype of neurogenic stuttering, it will not be used as a separate term in this thesis. Drug-induced stuttering is stuttering due to the use of medication and is sometimes seen as a subtype of neurogenic stuttering. However, the cause of stuttering is different from that in neurogenic stuttering and the stuttering can be stopped by discontinuing the

medication. Therefore, it will be considered as a subtype of acquired stuttering rather than a form of neurogenic stuttering (Van Borsel, 2014).

This thesis focusses on neurogenic stuttering. As most of the terms and knowledge about neurogenic stuttering is derived from knowledge about the developmental form of stuttering, characteristics and research of developmental stuttering will be described first in the following paragraphs.

2. Developmental stuttering

In the Diagnostic and Statistical Manual of Mental Disorders 5 (DSM-5), developmental stuttering is referred to as a 'childhood-onset fluency disorder'. To be diagnosed with this childhood-onset fluency disorder, the disturbed fluency in the speech has to have the onset of the disfluencies in the early developmental period and cause "anxiety about speaking or limitations in effective communication, social participation, or academic or occupational performance, individually or in any combination"(American Psychiatric Association, 2013, pp. 45 - 46). Additionally, there should be no other disorders or medical conditions that may cause the disfluencies (American Psychiatric Association, 2013). Most of the literature about stuttering also includes NSBs and anxiety as characteristics of stuttering, in addition to the speech characteristics. These characteristics will be discussed separately.

2.1 Speech characteristics

As stated above, the distinction between speech of normally fluent speakers (NFS) and people who stutter (PWS) is more complex than just the occurrence of disfluencies. Dysfluencies occur in speech of both NFS and PWS. However, some types of disfluencies do occur more frequently in stuttered speech. The classification of disfluency types that is used in the literature varies. The DSM-5 for example, describes the following disfluencies occurring in stuttering: "sound and syllable repetitions, sound prolongations of consonants as well as vowels, broken words (e.g. pauses within a word), audible or silent blocking (filled or unfilled pauses in speech and monosyllabic whole word repetitions, e.g. "I-I-I-I see him")" (American Psychiatric Association, 2013, pp. 45-46). Guitar (1998) also counts part-word repetitions, monosyllabic word repetitions, prolongations and blocks as disfluencies but adds successful avoidance behaviours to this list. In this thesis, the classification of disfluencies from Yairi and Seery (2015) will be adopted. These are similar to the disfluencies listed by the American Psychiatric Association (2013). The disfluency types more typical for stuttered speech are called stuttering-like disfluencies (SLDs) and include part-word repetition (sound or syllable repetitions), single-syllable word repetitions and dysrhythmic phonation (Yairi & Seery, 2015). Dysrhythmic phonation includes prolongations of sounds, blocks and tense pauses. In this thesis, no distinction is made between tense pauses and blocks, because tense pause has a low identification reliability (Yairi & Seery, 2015). The successful avoidance behaviours listed by Guitar (1998) as disfluencies will not be added to this list since these would be difficult to correctly identify.

Other disfluencies (ODs) are disfluencies that are typical for normally fluent speech. These disfluencies are multiple-syllable word repetitions, phrase repetitions, interjections and revisions or incomplete utterances.

Table 1 Classification of disfluency types, adopted from Yairi and Seery (2015, p. 92)

	Disfluency type	Description
Stuttering-like disfluencies (SLD)	Part-word repetition	Sound repetitions ("f-five") Syllable repetitions ("ba-baby")
	Single-syllable word repetitions	Repetition of words that consists of only one syllable ("but – but")
	Prolonged sounds	Audible elongations of sounds ("sssssome")
	Blocks	Articulators that are fixed in a certain position, blocking the air flow of speech ("ta-ble").
	Tense pause*	Breaks that occur between words*
Other disfluencies (OD)	Multiple-syllable word repetitions	Repetition of words that consist of more than one syllable ("happy – happy")
	Phrase repetitions	Repetition of a segment of a phrase longer than one word ("I was, I was").
	Interjections	Interruption of speech with sounds such as "um" and "uh".
	Revision/Abandoned utterance	Incomplete utterance, utterance that is changed mid-sentence.

*in this thesis coded also coded as 'blocks'

While SLDs are typical for stuttered speech, they can also occur in the speech of fluent speakers. To distinguish stuttered speech from fluent speech, it is advisable to gather estimates of the frequency of disfluencies in the average population. A disfluency rate of more than 1 standard deviation (SD) difference from the mean can be defined as abnormal (Logan, 2015). Several studies have been done to determine the amount of disfluencies in children and in adults with fluent speech. In children the average disfluency frequency (both SLDs and ODs) was 6.7 disfluencies per 100 words or 6.2 disfluencies per 100 syllables. In adults, the reported mean of disfluencies was four to seven disfluencies per 100 syllables (Logan, 2015). In her masters' thesis, Vanopdenbosch (2013) studied the amount of disfluencies of 24 Dutch-speaking older adults, these were classified into three age groups; four participants of 50-69 years old, ten of 70-79 years old and ten people of over 80 years old. She found an overall average of 4.3 disfluent words per 100 words. The youngest group, 50-69 years old had the least amount of disfluencies (3.0% versus 5.0% in 70-79 year olds and 4.2% in people over 80 years old). In all age groups and speech tasks, interjections were by far the largest group of disfluencies and accounted for 62-67% of all disfluencies. Repetitions of monosyllabic words and revisions came second and third, each accounting for 8-10% of all disfluencies. Blocks and word-finding difficulties (disfluency where the speaker expresses trouble recalling the right word) occurred the least with 0.5% and 0.3%. (Vanopdenbosch, 2013).

In the above mentioned studies, both SLDs and ODs were measured in people who do not stutter. In people who stutter, the frequency of SLDs is higher. Silverman and Zimmer (1979)

found an average of 7.9 SLDs per 100 words in adult males who stutter and 12.1 SLDs per 100 words in adult females who stutter in spontaneous speech. The total amount of disfluencies per 100 words was 17.3 for adult men and 20.5 for adult woman who stuttered.

To distinguish stuttered speech from non-stuttered speech, Guitar (1998), suggests a criterion of 10% disfluencies (ODs and SLDs) to distinguish stuttered speech from fluent speech. However, the contrast between the frequency of SLDs in NFS and PWS is greater than the difference in amount of ODs. In a normative study, Ambrose and Yairi (1999) recorded 144 preschool children, of which 90 exhibited stuttering. The control group of fluent speakers had on average 1.3 SLDs per 100 syllables with a standard deviation of .83. To diagnose disorders, often a criterion of differing more than two standard deviation from the mean is used. Based on the normative study of developmental stuttering, this would mean displaying more than 3 SLDs per 100 syllables. Therefore, a lot of studies handle a threshold of 3% SLDs as a cut-off to diagnose stuttering (Yairi & Seery, 2015).

2.2 Non-speech behaviours

In addition to speech disfluencies, PWS often display involuntary movements. Frequently observed non-speech movements include eye blinking, grimacing, sudden exhalations of breath (Bloodstein & Ratner, 2008). The movements are involuntary and can occur in any part of the voluntary musculature of the body. They often appear during stutters but can also be recognized during speech without observable disfluencies (Conture & Kelly, 1991). The Stuttering Severity Instrument – 4 (SSI-4) is a diagnostic instrument for determining stuttering severity. It includes measurements of stuttering frequency, stuttering durations and NSBs. In the SSI-4, the NSBs, called *physical concomitants,* are divided into four groups: distracting sounds, facial grimaces, head movements and movements of the extremities. They are evaluated on how distracting and noticeable the movements are on a 6-point scale (0 = none, 1 = not noticeable unless looking for it, 2 = barely noticeably to casual observer, 3 = distracting, 4 = very distracting, 5 = severe and painful looking) (Riley, 2009).

Non-speech behaviours are often seen as a persons' reaction to their stutters. Van Riper (1971) describes what he calls "Dr. Jekyll stutterers": who have severe avoidance behaviour of stuttering. When confronted with a stutter, the person tries to deny or disguise the stutter by looking away or shutting their eyes. "Mr. Hyde stutterers" on the other hand "suffer visibly, almost revel in their verbal misery" and show non-speech behaviours of struggle and facial contortions (Van Riper, 1971, p. 203). According to Van Riper (1971), these avoidance reactions are the result of a low body-image. Guitar (1998) describes speech disfluency behaviours as the "core-behaviours" of stuttering and the non-speech behaviours as 'secondary behaviours' to the stuttering. He distinguishes two behavioural reactions to stutters: escape behaviours and avoidance behaviours. The non-speech behaviours like eye blinks and grimacing are, according to Guitar (1998) a strategy to escape or avoid stutters. Guitar (1998) describes several phases of developmental stuttering: normal disfluency, borderline stuttering, beginning stuttering, intermediate stuttering and advanced stuttering. According to him, non-speech behaviours usually only appear from the beginning stuttering phase, because the child begins to react on the stutters. From this moment on, the child begins to have escape and avoidance behaviour more frequently. The escape behaviours are motor movements in order to escape a stutter while avoidance

behaviours can be substitutions of words, circumlocutions or postponements. By the time the child reaches the advanced stuttering phase, avoidance of stutters is the most extensive non-speech behaviour (Guitar, 1998). In the theories of Van Riper (1971) and Guitar (1998), non-speech behaviours develop as a way to avoid or get out of stutters. These theories were believed by researchers for a long time (Yairi & Seery, 2015). However, several studies report physical movements that are present early in the development of stuttering (Conture & Kelly, 1991; Schwartz, Zebrowski, & Conture, 1990; Yairi & Ambrose, 2005). In a study of Schwartz et al. (1990) the relationship between non-speech behaviours and time since stuttering onset was examined. The time between data collection and onset of stuttering ranged between one to twelve months. They hypothesised that all children who stutter display non-speech behaviours regardless of time after onset of stuttering. Their findings supported this hypothesis: all participating children exhibited non-speech behaviours. Yairi and Ambrose (2005) reported that 53% of the parents reported that their children displayed non-speech behaviours at onset of stuttering. Conture and Kelly (1991) found that young stutterers (3 – 7 years old) exhibited 1.5 non-speech behaviours on average during stutters. However, there was no information available about the time between onset of stuttering and data collection in this study. In another study, Yairi, Ambrose, and Niermann (1993) examined the speech and facial and head movements of children 1 – 12 weeks after stuttering onset. They analysed the facial and head movements during ten SLDs per child and found a mean of 3.2 facial- or head movements per disfluency. The amount of movements had declined in a 3 month-follow up (2.4 movements per disfluency) and a 6-month follow up (1.9 movements per disfluency). The amount of SLDs also declined over time with a steeper slope than the facial and head movements. This suggests that stuttered speech can already be complex at onset of stuttering (Yairi et al., 1993). These studies contradict the widely believed theory that non-speech movements develop over time as a reaction to stuttering.

Different terms are used in the literature to describe non-speech behaviours, such as *secondary behaviours* (Guitar, 1998), *associated movements* (Bloodstein & Ratner, 2008*) and physical concomitants* (Riley, 2009). The most frequently used term, *secondary behaviours*, suggests that non-speech behaviours are a consequence of stuttering. The other terms also suggest a dichotomy in stuttering behaviours with the non-speech behaviours inferior to the speech behaviours of stuttering. The hypothesis that non-speech behaviours are secondary to the speech behaviours of stuttering is debatable, as children are known to display non-speech behaviours right at the onset of stuttering (Conture & Kelly, 1991; Schwartz et al., 1990; Yairi & Ambrose, 2005). It is therefore possible that non-speech behaviours are an intrinsic symptom of stuttering, rather than a reaction to the stuttering or a secondary symptom (Logan, 2015; Yairi & Seery, 2015). That is why in this thesis, the neutral term *non-speech behaviours* (NSB) is used.

2.3 Anxiety

It is often reported that people with developmental stuttering develop certain emotions towards their stuttering and speech. The emotions that can accompany stuttering include “fear, dread, anxiety, being trapped, panic, embarrassment, shame, humiliation, anger, resentment, and other unpleasant feelings” (Yairi & Seery, 2015, p. 120). These emotions can occur before stutters, during stuttering and after the stutter. Anxiety is one of the emotions that is often reported in literature about stuttering. There are two types of anxiety: trait anxiety and state anxiety. The

inherent level of anxiety is the trait anxiety and state anxiety can be described as the anxiety specific to a situation or condition (Kefalianos, Onslow, Block, Menzies, & Reilly, 2012). Young children who begin to stutter often do not react to their stutters. However, after some time the child will become aware of the unpleasantness of stuttering and will begin to react to the stutters. These reaction of the person who stutters to their own stutters and the reaction of other people to the stuttered speech can influence the development of negative emotions towards stuttering (Yairi & Seery, 2015). The feelings and attitudes towards stuttering can be a big part of the stuttering problem (Guitar, 1998). Guitar (1998) describes that these feelings and attitudes of a person who stutters, including frustration and embarrassment, can influence stuttered speech behaviour. On the other hand he states that anxiety is not a distinctive characteristic of stuttering. In the DSM-5, anxiety about speaking or limitations in communication, social participations or performance were added as diagnostic criteria, thus stating that anxiety or social consequences are always present with developmental stuttering (American Psychiatric Association, 2013).

Messenger, Onslow, Packman, and Menzies (2004) found that adults who stuttered more often expected negative social evaluation in social contexts than adults who did not stutter. In non-social situations, PWS did not differ from NFS in amount of expected negative evaluation. In a study of Kraaimaat, Vanryckeghem, and Van Dam-Baggen (2002) the levels of social anxiety of stuttering adults were examined using a questionnaire. Adults who stuttered scored significantly higher on 'emotional tension in social situations' compared to non-stuttering adults. However, not all stuttering adults experienced high levels of anxiety. In another study Ezrati-Vinacour and Levin (2004) examined levels of trait anxiety and stuttering severity in 47 adult males with stuttering and 47 fluent speaking males. Overall, the persons who stuttered had a higher level of trait anxiety than persons without stuttering. The level of trait anxiety did not differentiate between levels of stuttering severity, so higher trait anxiety was present across all levels of stuttering severity. Anxiety in social communication however, was higher among persons with severe stuttering compared to persons with mild stuttering.

Iverach et al. (2009) examined the prevalence of anxiety disorders in adults with stuttering. They found that anxiety disorders were much more common in adults who stutter (27.2 %) than in the control group (5.3%). Social phobia was most often diagnosed in the stuttering group with a 21.7% prevalence. In a meta-analysis, Craig and Tran (2014) analysed results from nineteen studies. Eleven of these studies assessed trait anxiety and eight assessed social anxiety. They found that adults who stutter have a moderately higher trait anxiety but a substantially elevated social anxiety (Craig & Tran, 2014). The effect sizes they found were moderate for trait anxiety and high for social anxiety. Two hypotheses exist about the relationship between stuttering and anxiety. The first that anxiety is a trait of PWS, the second that the anxiety frequently found in PWS is caused by the stuttering. Alm (2014) found in a review that no studies found shyness, social anxiety or similar traits to appear more frequent in pre-schoolers who stutter than in children who do not stutter. This indicates that social phobia and anxiety is developed as a result from stuttering (Alm, 2014).

3. Neurogenic stuttering

Neurogenic stuttering is often described as a rare condition, yet many clinicians have encountered patients with neurogenic stuttering. One fourth of the clinicians who participated in a survey in Belgium said to have recently worked with at least one patient with neurogenic stuttering (Theys, van Wieringen, & De Nil, 2008). In another study, 319 stroke patients were screened for speech and language problems by a speech- and language pathologist. Of these patients, seventeen were diagnosed with neurogenic stuttering, resulting in an incidence rate estimation of 5.3%. The neurogenic stuttering persisted in eight patients for at least six months, which resulted in a prevalence estimate of 2.5% (Theys, van Wieringen, Sunaert, Thijs, & De Nil, 2011). Similarly to developmental stuttering, the available information on the speech characteristics and NSBs of persons with neurogenic stuttering will be discussed.

3.1 Speech characteristics

The diagnosis neurogenic stuttering is based on the occurrence of 3% or more stutter-like disfluencies during speech and an onset during adulthood linked to neurological damage. Neurogenic stuttering can co-occur with other speech- and language disorders like aphasia, dysarthria or apraxia of speech (Theys et al., 2011). It is important to note that these other speech- and language disorders can also cause disfluencies, so to diagnose neurogenic stuttering it is important to differentiate between disfluencies caused by other speech- and language disorders. Other disfluencies, revisions and interjections for example, can be caused by word-finding difficulties as a result of aphasia (Papathanasiou, Coppens, & Potagas, 2013). In neurogenic stuttering however, the speaker knows what they want to say, but has trouble saying the words (Yairi & Seery, 2015).

Some authors argued that the speech characteristics of neurogenic stuttering are different from the speech characteristics seen in developmental stuttering. Canter (1971) named several characteristics that differentiate developmental stuttering from neurogenic stuttering. He proposed seven diagnostic criteria to help the clinician identify neurogenic stuttering. Helm-Estabrooks (1999) reviewed and revised the characteristics proposed by Canter (1971) which resulted in the following characteristics (Helm-Estabrooks, 1999, p. 260):

1. "Dysfluencies occur on grammatical words nearly as frequently as on substantive words.
2. The speaker may be annoyed but does not appear anxious.
3. Repetitions, prolongations, and blocks do not occur only on initial syllables of words and utterances.
4. Secondary symptoms such as facial grimacing, eye blinking, or fist clenching are not associated with moments of dysfluency.
5. There is no adaptation effect.
6. Stuttering occurs relatively consistently across various types of speech tasks."

These characteristics are still cited in a lot of literature to this day. Helm-Estabrooks describes neurogenic stuttering as "stuttering associated with acquired neurological disorders (SAAND)". According to her it is important that aphasia is ruled out before diagnosing SAAND.

Contrarily to what Canter (1971) and Helm-Estabrooks (1999) suggest, there is evidence that the speech characteristics of neurogenic stuttering are not that different from the speech characteristics of developmental stuttering. Making the distinction between developmental stuttering and neurogenic stuttering purely on speech characteristics appears to be difficult for professionals. In a study of Van Borsel and Taillieu (2001), nine speech and language pathologists were asked to place patients in a developmental stuttering group or a neurogenic stuttering group on the basis of a three minute speech sample. Four patients had developmental stuttering diagnoses and the other four were diagnosed with neurogenic stuttering. The classification of the patients by the participating speech and language pathologists was wrong in 24%, and in 42% of the cases the speech and language pathologist was not sure of his/her judgement. Of the eight patients, two developmental stutterers were diagnosed correctly by all speech and language pathologists (SLP). The amount of blocks and non-speech behaviours were mentioned as reasons for the classification of developmental stuttering. Only one neurogenic stuttering patient was correctly identified by all SLP's and in this case word finding difficulties were one of the main reasons for the correct diagnoses. The amount of misjudgements on the basis of symptomatology and the uncertainty of the SLP's could mean that there are a lot more similarities in stuttering behaviour between developmental stuttering and neurogenic stuttering than the diagnostic criteria of Canter (1971) and Helm-Estabrooks (1999) suggest. The six features of Helm-Estabrooks (1999) could therefore be used as a 'rule of thumb' instead of strict diagnostic rules (Lundgren, Helm-Estabrooks, & Klein, 2010).

In most cases, however, the distinction between developmental stuttering and neurogenic stuttering is not difficult. Neurogenic stuttering usually has an onset in adulthood, after some form of neurological damage. In contrast, developmental stuttering has an onset during childhood in children with no history of neurological problems. It is therefore better to base the distinction between developmental stuttering and neurogenic stuttering purely on whether or not neurological damage has occurred before stuttering onset instead of on speech characteristics.

The distinction between neurogenic and psychogenic stuttering is more difficult however. Both types of stuttering generally have an onset in adulthood. Stuttering after an emotional trauma and no clear neurological causes, could be identified as psychogenic stuttering. Stuttering after neurological trauma can both be attributed to the neurological damage and to the emotional stress caused by the neurological trauma. In some cases it might be impossible to know whether the underlying cause of stuttering is neurological or psychogenic (Helm-Estabrooks & Holz, 1998). For example, in the articles of Attanasio (1987), Nowack and Stone (1987) and Theys, van Wieringen, Tuyls, and de Nil (2009), reporting a case of neurogenic stuttering, psychogenic stuttering cannot be ruled out. Interestingly, the cases of Attanasio (1987) and Nowack and Stone (1987) were described as anxious. The 36-year-old male described by Attanasio (1987) began stuttering after marital problems, the stuttering became worse when the problems worsened and became most severe during divorce. Even though the described subject was convinced that the stuttering was linked to the marital problems, Attanasio (1987) suggests that the stuttering might be linked to his epilepsy rather than to his psychological stress. In this case, however, psychogenic stuttering does seem more likely than neurogenic stuttering, as psychological stress

can be linked to the onset of stuttering whereas no epileptic attack had occurred shortly before onset of stuttering. This provides stronger evidence towards psychogenic stuttering than neurogenic stuttering. In the case described by Nowack and Stone (1987), the possibility that there is a psychological component to the stuttering is mentioned, although this possibility is denied by the husband of the subject. The subject had recently moved and lost a job before stuttering onset, as well as cerebral infarction. A speech pathologist concluded that the stuttering most likely had a neurological basis, but where this conclusion is based on is not mentioned in the article. In the study of Theys et al. (2009), the 16-year-old boy described in the case study did have neurological symptoms suggestive of cerebellar encephalitis but this could not be diagnosed on the basis of medical examination. A psychiatric evaluation did not lead to a psychiatric diagnosis but according to the psychiatrist a psychological factor could not be ruled out.

3.2 Non-speech behaviours and anxiety

Both Canter (1971) and Helm-Estabrooks (1999) state that non-speech behaviours are not present in people with neurogenic stuttering and that people with neurogenic stuttering are not anxious about their speech. This has been adopted in a lot of literature. Ashurst and Wasson (2011) and Prasse and Kikano (2008), for example, both state that neurogenic stuttering is easily differentiated from developmental stuttering because people with neurogenic stuttering usually do not display anxiety about talking and the stuttering is not accompanied by non-speech behaviours (NSB).

Ringo and Dietrich (1995) studied all characteristics described by Helm-Estabrooks (1999) and Canter (1971). They read thirty articles with a total of 79 described cases of neurogenic stuttering and investigated whether the characteristics applied to the described cases. Some characteristics were found in a majority of the described cases, although not all cases reported all characteristics. Emotional response, for example, was described in 41 of the 79 cases. Of these 41 cases, 80% did not feel anxiety about their speech. Likewise, the existence or absence of NSBs was mentioned in 53 cases, of which 70% reported no NSBs. In a survey study of Theys, van Wieringen, and De Nil (2008), questioning speech and language therapists about cases of neurogenic stuttering they had seen, a total of 58 cases were described. Of these cases, a total of 32 (55%) cases were reported to have NSBs such as facial grimaces, associated limb movements, postponement behaviours and avoidance behaviours. Emotional reactions to their stuttering were seen in 37 patients, e.g. frustration, irritation, fear, crying and anger. These results do not support the findings of Ringo and Dietrich (1995) or the characteristics proposed by Canter (1971) and Helm-Estabrooks (1999) as they report NSBs and emotional reactions to stuttering in over half of the cases.

Since the study of Ringo and Dietrich (1995), more case-studies of neurogenic stuttering have been published. For this thesis, a total of 60 articles, describing a total of 86 cases of neurogenic stuttering, have been reviewed. Some of these studies were also reported in the study of Ringo and Dietrich (1995). There were eighteen articles reporting on cases of neurogenic stuttering that

possibly reported on anxiety and/or NSBs, but were not available in full-text. They were referenced by other articles, but not in detail, so they are not included.

Of the 21 cases of whom emotional reactions were described, just over half (61%) were reported to show some emotional reactions about their speech. The reported reactions varied, for example mild annoyance (Koller, 1983) and feeling miserable and afraid of speaking (Bijleveld, Lebrun, & van Dongen, 1994). Only two cases were literally described as anxious about their speech (Attanasio, 1987; Nowack & Stone, 1987). Another eight cases did not show any anxiety about the stuttering. However, interpretation of these results has to be done with caution, as in 75% of the cases of neurogenic stuttering reported in the literature, no information is given about emotions or anxiety about the stuttering.

NSBs were reported in 46 of the 86 cases (53%). Out of these 46 cases, NSBs were seen in nineteen cases, for example involuntary blinking (Lebrun, Bijleveld, & Rousseau, 1990) and grimacing (Sahin, Krespi, Yilmaz, & Coban, 2005; Tani & Sakai, 2010). In some cases only one NSB was reported, while in other cases up to seven NSBs were mentioned. Four articles mentioned a change in NSBs over time, in three of those cases the amount of NSBs increased over time (Lebrun, Rétif, & Kaiseer, 1983; Stewart & Grantham, 1993; Vanhoutte et al., 2014). In the fourth case however, there were less reported NSBs in the 3rd test moment than in the first two test moments as his fluency improved. Strikingly, this case-study reported the presence of NSBs even though the speech was seemingly fluent (Theys et al., 2009). Of the 46 cases, 28 cases were reported to have no NSBs. Of the remaining 40 cases (47%) NSBs were not reported in the article. An overview of all cases with reported emotional reactions and/or NSBs is given in table 2.

Most of the case-studies about neurogenic stuttering do not report levels of anxiety and only a little over half of the studies report NSBs. The studies that do report on NSBs, do not support the characteristics of Canter (1971) and Helm-Estabrooks (1999) as the occurrence of NSBs was reported in 40% of the cases. The occurrence of NSBs found in the literature is less than the reported 55% in the survey study of Theys et al. (2008), but more than the 30% reported in the study of Ringo and Dietrich (1995).

As most of the studies are focussed on describing the speech characteristics of stuttering, and report anxiety or NSBs based on observations instead of tests or questionnaires, comparing different studies is complicated. Vanhoutte et al. (2014) for example reported that the PWS was "concerned" about their stuttering, Lebrun and Leleux (1985) describe that the PWS was "complaining about their stuttering" while Stewart and Grantham (1993) report "embarrassment, anger and overwhelming hatred". Although these are all clear emotional reactions to stuttering, they are not objective descriptions of anxiety levels. It is difficult to extract from these observations which cases have mild emotional reactions to stuttering and which cases are experiencing anxiety about their stuttering. It is possible that some of the cases were falsely reported to have no anxiety about their stuttering because they did not show their anxiety. It also seems that the authors do not have the same definition of anxiety or NSBs. Leder (1996) for

instance, did report his case to have no NSBs. He also reported increased pitch and tension in shoulder and neck areas, which would fall under the definition of NSBs in this thesis. Bijleveld et al. (1994) also reports that NSBs were absent in her subject but at the same time reports nodding, facial tension, groping, starting, gasping and involuntary pitch changes. It could be that some articles report no NSBs because the definition of non-speech behaviours is different. It might even be that, because of the well-known characteristics of Helm-Estabrooks (1999), researchers do not expect non-speech behaviours and therefore do not look for them or report them.

Table 2, Overview of cases of neurogenic stuttering in the literature with reported emotional reactions and/or non-speech behaviours

Authors (year)	Age	Gender	Aetiology	Reported emotional reaction	Reported non-speech behaviours
Attanasio (1987)	36	Male	Epilepsy, possible psychogenic factors	Anxiety and concern	Tension
Bijleveld et al. (1994)	65	Female	Stroke(s)	Felt miserable, afraid of speaking, self-confidence was diminished.	Nodding, facial tension but no grimacing, groping, starting, gasping, involuntary pitch changes.
Carluer et al. (2000)	58	Male	Stroke	Considered stuttering a disabling social symptom	None
Heuer, Sataloft, Mandel, and Trayers (1996)	40	female	Moya Moya disease	-	Jaw tension
Heuer et al. (1996)	53	male	Stroke	-	Aversion of eye gaze, decreased speech volume, eye closing
Koller (1983)	55	Male	Parkinson's disease	Slightly annoyed	None
Koller (1983)	62	Male	Parkinsonism	Mild frustration	None
Koller (1983)	65	Male	Parkinsonian symptoms	Mild annoyance	None
Lebrun et al. (1983)	59	Male	Parkinsonism	-	Closed eyes synkinetically during blocks and prolongations
Lebrun et al. (1990)	23	Male	Penetrating brain lesion	Aware but not desperate	Involuntary blinking, closing of the eyes, slight bending of the head
Lebrun, Leleux, and Retif (1987)	61	Male	Stroke	-	Constant eye blinking
Leder (1996)	29	Male	Parkinson	No word fears or avoidances, no situational fears, very distressed about stuttering.	No non-speech behaviours, observational tension in shoulder and neck areeas, increased pitch.
Mowrer and Younts (2001)	36	Male	Multiple sclerosis	Disturbed by the repetitions	None
Nowack and Stone (1987)	55	Female	Right-hemisphere cerebral infarction, stuttering onset after series of psychological stresses.	Annoyed and anxious.	Did not show poor eye contact, distracting sounds or excessive body movements.
Rosenbek, Messert, Collins, and Wertz (1978)	53	Male	Stroke	-	Increased effort, eye blinking, grimacing
Rosenbek et al. (1978)	52	Male	Stroke	-	Increased effort, eye blinking, grimacing
Rosenbek et al. (1978)	61	Male	Stroke	-	Increased effort
Rosenbek et al. (1978)	54	Male	Strokes	-	Increased effort
Rosenbek et al. (1978)	65	Male	Stroke	-	Increased effort, eye blinking, grimacing

Rousey, Arjunan, and Rousey (1986)	41	male	Closed head injury	Great deal of fear and avoidance of speaking situation	-
Sahin et al. (2005)	65	Female	Stroke	-	Facial grimcing, eye-blinking, fist clenching, lip tremor
Stewart & Grantham (1993)	21	Female	Migraine and bilateral tremor	Anger, hatred of the way she spoke, frustration	Loss of eye contact
Tani and Sakai (2010)	45	Male	Stroke	-	Closing of the eyes, increased tonus of facial muscles, grimacing
Theys et al. (2009)	16	Male	Rotavirus infection, suggestive cerebellar encephalitis	Annoyed	One month after onset: Played with hand, eye squinting, tense muscles in face and forward head movement. Three months after onset: lifting eyebrows, turning eyes upward, watching hands, careful and slow speech. One year, three months after onset: revisions, lifting eyebrows, eyes turned upwards, looking away.
Van Borsel, van Lierde, van Cauwenberge, Guldemont, and van Orshoven (1998)	69	Male	TBI	-	Facial grimacing
Vanhoutte et al. (2014)	28	Female	Multiple strokes, neurological surgery.	Concerned.	Slight increase of physical concomitants, nodding of the head and frowning.

4. Research questions and hypotheses

In short, a lot of information about anxiety and non-speech behaviours in neurogenic stuttering is missing in the literature as many case-studies do not explicitly report on presence or absence of these behaviours. The information that is available on anxiety and NSBs is largely based on 22 articles that do report on NSBs and eighteen articles that reported on anxiety in neurogenic stuttering. Of these studies reporting on emotional reactions, more than half of the cases do have emotional reactions to their stuttering although the severity of these reactions varies. In 40% of the cases that reported on NSBs, NSBs occurred during speech. However, a survey study by Theys et al. (2008) reported that NSBs occurred in 55% of the reported patients with neurogenic stuttering. These findings suggest that the characteristics of neurogenic stuttering proposed by Canter (1971) and Helm-Estabrooks (1999) are not conclusive. In this thesis a larger group of people with neurogenic stuttering was analysed to get a better view on the characteristics of neurogenic stuttering and the non-speech behaviours in particular. Therefore, the following research questions and hypotheses form the basis of this research.

1. *Do people with neurogenic stuttering present with more, longer and/or severe non-speech behaviours when compared to fluent speakers?*

The literature about the occurrence of non-speech behaviours in people with neurogenic stuttering is inconsistent. Therefore, the expectation is that the presence of non-speech behaviours will vary between participants. Overall, the hypothesis is that on average, people with neurogenic stuttering will present with more non-speech behaviours than fluent speakers. Secondly, it is possible that the NSBs of people with neurogenic stuttering will be longer in duration. It is also expected that the NSBs of people with neurogenic stuttering will be more distracting and thus more severe.

2. *Is the stuttering frequency (proportion of SLDs) a predictor of the amount, duration and severity of non-speech behaviours in speakers?*

In the case-study of Rosenbek et al. (1978) the three subjects with the severest neurogenic stuttering, i.e. the highest proportion of SLDs, also had the most non-speech behaviours. In other studies this relationship is not mentioned, as non-speech behaviours are almost always only described and not counted or measured for duration or severity. In studies on NSBs in developmental stuttering, these NSBs are measured per SLD (Conture & Kelly, 1991; Yairi et al., 1993). If NSBs mostly occur during instances of SLD, it would be expected that a higher proportion of SLDs results in a higher amount of NSBs. The expectation is that stuttering frequency is also correlated to severity and duration of NSBs.

3. *What are predictors of the amount, duration and severity of non-speech behaviours in people with neurogenic stuttering?*

It is expected that there will be variation within the group of participants with neurogenic stuttering in the proportion, duration and severity of NSBs. This question will address what factors might be a predictor of the amount, severity and duration of non-speech behaviours of people with neurogenic stuttering. The first one is the amount of both SLDs and NDs. People

who present with severe stuttering might also have a higher frequency and more severe or longer non-speech behaviours. One theory about non-speech behaviours, is that they develop over time as a reaction to the stutters (Guitar, 1998). This suggest that time post-onset can be a predictor, since a longer time post-onset results in more time in which non-speech behaviours could have been developed. Secondly, if non-speech behaviours develop as a reaction to stutters, the emotions and attitudes towards stuttering can be a predictor of non-speech behaviours. It may be that people who are more anxious about their speech, react more to their stuttering and thus develop a higher frequency, longer or more severe, non-speech behaviours. It is also possible that the presence of co-occurring disorders, like aphasia, dysarthria of apraxia of speech, result in more non-speech behaviours because the person has more trouble speaking. Lastly, there is evidence that speech characteristics of people with neurogenic stuttering differ between aetiologies (Theys et al., 2008). Therefore the aetiology may be a possible predictor of NSBs.

5. Method

5.1 Participants

The participants consisted of 27 Dutch-speaking persons, 9 females and 18 males, aged 46 - 86 (M: 66.57 , SD = 12.01) with a diagnosis of neurogenic stuttering based on having more than 3% SLDs during one or more speech tasks. Of the 27 participants, nineteen were reported on in the stroke study by Theys et al. (2011). Other participants were referred to Theys for assessment in light of possible neurogenic stuttering following various aetiologies.

Twenty-three of the participants started stuttering after a cardiovascular arrest (CVA), two had a traumatic brain injury (TBI), one had amyotrophic lateral sclerosis (ALS) and one had Parkinson's disease (PD). One participant had a diagnosis of dementia, next to the CVA. Of the participants, fifteen were known to be diagnosed with co-occurring speech and/or language disorders such as aphasia (10 participants), dysarthria (10 participants) and apraxia of speech (2 participants). Of the fifteen participants with co-occurring speech and/or language disorders, 6 participants suffered from two or more co-occurring disorders. Time post stuttering onset varied from two days to over 3,5 years (M = 76.5 days, SD = 281).

The control group consisted of 20 elderly persons, 10 females and 10 males, aged 71 to 92 (M = 80.29, SD = 6.53). None of the participants in the control group had neurological disorders, speech- or language disorders or took any medication that could affect speech, language or memory. Similar to the neurogenic stuttering participants, all control participants were native Dutch speakers.

An overview of all participants and their age, gender and for the neurogenic stuttering group the medical aetiology and time post-onset, can be found in appendix A.

5.2 Recordings

The participants in the neurogenic stuttering group completed between 1 and 3 test-sessions over one year. In this thesis only the recording of the first test session was used. This test session took place as soon as possible after stuttering onset. The test-sessions consisted of speech- and

language tests, reading-tasks and a conversation. All test-sessions were video recorded. Only the recordings of the conversations were analysed in this thesis. The conversation topics included work or hobbies and varied between participants based on their interests.

The control group was tested by Vanopdenbosch (2013). The test-session included a revised Boston Naming Test (Mariën, Mampaey, Vaervaet, Saerens, & De Deyn, 1998), a reading task, describing pictures and a conversation of approximately 15 minutes about a subject of interest to the participants. Again, only the recordings of the conversations were used in this thesis.

5.3 Transcriptions

For the neurogenic stuttering group, a 300-word conversation sample from the middle of a conversation was selected and transcribed. Selecting the middle of the conversation should minimise any effect of possible unease or uncomfortable feelings at the beginning of the conversation or tiredness at the end. Repeated words and interjections were not included in the word count, to ensure the 300-word sample consisted only of meaningful speech (Yaruss, 1998). When it was not possible to extract a 300-word sample, a minimum of 200 words was considered necessary for a speech sample to be included in the analysis. For four participants (NS3, NS4, NS10 and NS15) the recorded conversation consisted of 200 - 299 words. Five other neurogenic stuttering participants (NS23, NS24, NS25, NS26 and NS27) and three participants of the control group (C3, C5 and C10) were excluded from the analysis because the speech sample was smaller than 200 words.

The videos were played repeatedly until the researcher was confident the transcription was correct. If the researcher could not identify what a participant said, this part was transcribed as ‘unintelligible’ and excluded from analysis. For thirteen participants a transcription made by Theys et al. (2011) was available. These transcriptions were used if they consisted of a 300-word sample from the middle of a conversation. The transcriptions were checked and, if necessary, adjusted.

For the control-group, 300-word samples from the middle of the conversation were transcribed by Vanopdenbosch (2013). These videos and transcriptions were provided by Vanopdenbosch and used in this study. All transcriptions were checked and adjusted if necessary.

5.4 Analysis of speech and non-speech behaviours

For the analysis of the speech and non-speech behaviours, all stuttering-like disfluencies (SLDs), other disfluencies (ODs), non-speech behaviours (NSBs) and gestures present during the conversation samples were marked using ELAN (Max Planck Institute for Psycholinguistics, 2017). An overview of the tiers used for coding is presented in table 4. The classification of speech disfluency behaviours was based on Yairi and Seery (2015).

The classification of non-speech behaviours was adopted from the SSI-4 (Riley, 2009). Because non-speech behaviours can also occur during seemingly fluent moments, all observed behaviours and movements during speech were annotated (Conture & Kelly, 1991; Theys et al., 2009). Leg and foot movement was not measured because this was not visible in the videos. In two participants of the control group, only the head and shoulders were visible. The annotation

'poor eye contact' was retracted from analysis, because the communication partner was not visible in most recordings and it was therefore not possible to determine reliably whether eye contact was established.

All annotated non-speech behaviours were scored according to the five point scale of the SSI-4 (1 = not noticeable unless looking for it, 2 = barely noticeably to casual observer, 3 = distracting, 4 = very distracting, 5 = severe and painful looking) (Riley, 2009). This five point scale is designed to only score the severity of non-speech behaviours associated with stuttering. In this study, all movements were annotated and scored, and therefore the scale was altered to make it more suitable to score all movements. This adjusted scale can be seen in table 3.

Table 3 Used five point scale to score severity of non-speech behaviours

Scale	Description
1	Not noticeable movement unless looking for it, non-distracting movements that fit context and/or natural speech.
2	Movements that attract a little bit of attention, for example abrupt movements or movements that do not entirely fit context.
3	Distracting movements, movements with noticeable tension
4	Very distracting movements, obvious tension in movement
5	Severely distracting and painful looking movements

Using this coding system for each conversational sample, data on frequency of occurrence and duration was obtained for all the ODs, SLDs and NSBs. For the latter, a severity score was also obtained.

Lastly, the amount of NSBs per SLD and NSBs per OD were obtained. Using the overlapping annotation function in ELAN, all NSBs that overlapped with SLDs or ODs were counted. Then an average of NSBs per SLD and NSBs per OD per participant was calculated.

Table 4 Overview of tiers used in ELAN with descriptions. The classification of disfluencies is based on Yairi and Seery (2015) and classification of NSBs based on Riley (2009)

Behaviours			Description
Other disfluencies	Revision		Modification of a phrase, "I was baking – cooking"
	Interjection		Interjection of a sound such as "uh" or "uhm" between words
	Whole word repetition		Repetitions of words containing multiple syllables, "water water"
	Part phrase repetition		Repetition of multiple words within a phrase, "I want-I want"
Stutter behaviours	Block		Keeping the articulators in a fixed position. Blocks can be silent or have minimal sound, "-pasta", "pas-ta"
	Prolongation		Elongation of a sound, "wwwwwater"
	Repetition	Sound repetition	Repetitions of sounds, "b-baby"

		Syllable repetition	Repetitions of syllables, “wa-water”
		Single syllable word repetition	Repetitions of words that contain only one syllable, “I-I-I”
Non-speech behaviours	Distracting sounds	Noisy breathing	Audible breathing
		Whistling	Producing whistling sounds
		Sniffing	Audible and quick breathing in through the nose
		Blowing	Forcing air outward through rounded lips
		Clicking Sounds	Producing clicking sounds
		Other	Other sounds that do not meet criteria of any of the described distracting sounds
	Facial grimaces	Jaw jerking	Sudden and quick movement of the jaw
		Tongue protruding	Outward movement of the tongue
		Lip pressing	Tense closing of the lips
		Jaw muscles tense	Visible tensing of the jaw muscles
		Frowning	Frowning not related to spoken context
		Other	Other (tense) facial movements that do not meet criteria for any facial grimaces
	Head movements	Back	Head movement in backwards direction of speaker
		Forward	Head movement in forward direction of speaker
		Turning away	Head movement turning to side
		Poor eye contact *	Poor eye contact
		Constant looking around	Constant head and/or eye movement to different directions
		Other	Other head movements that do not meet criteria of any of the described head movements
	Movements of the extremities	Arm	Movement of the arm
		Hand	Movement of the hand
		Hands about face	Movement of the hands around the face of the speaker
		Torso	Movement of the torso
		Leg *	Movement of the leg
		Foot-tapping *	(Repetitive) up and down movement of the foot
		Swinging	Repetitive swinging to sides or back and forth of any of the extremities
		Other	Other movement of the extremities that do not meet criteria of any of the described movements
Gestures			Gestures that add to semantic and/or prosodic context

* Not included in the analysis

5.6 Predictors of non-speech behaviours

The hypothesised predictors of non-speech behaviours within the neurogenic stuttering group were frequency of SLD and OD, time post-onset, amount of co-occurring speech- and language disorders, medical aetiology and emotions and attitudes about stuttering. The co-occurring disorders were tested by Theys. Time post-onset and medical aetiology were obtained from the medical history of the participants. Because the different aetiologies were very unequally divided within the neurogenic stuttering group, it was not included as a predictor, as eighteen participants had the medical aetiology CVA, and only two had TBI, one ALS and one PD. The emotions and attitudes regarding stuttering, were based on scores of several questionnaires. All participants of the neurogenic stuttering group were asked by Theys to complete the Behaviour Assesment Battery for adults who stutter (Brutten & Vanryckeghem, 2003). This battery consists of four questionnaires about emotions and attitudes towards their speech: the speech situation checklist part 1: emotional reactions (SSC-ER), the speech situation checklist part 2: speech disorder (SSC-SD), the behaviour checklist (BCL) and Erickson's scale of communication attitudes (S-scale). In all questionnaires, a cut-off score of two standard deviations higher than the mean of fluent speakers was used to determine whether a score indicates severe emotions and/or attitudes.

The SSC-ER measures the amount of negative emotions that occur in speech situations. A total of 51 speech situations are listed, for example "speaking on the phone" and "making an appointment", and the participant has to score the amount of negative emotions they feel in these speech situations on a five point scale with one being "no negative emotions" and five being "a lot of negative emotions". A score over 125.71 on the SSC-ER indicates that the participant had severe negative emotional reactions to speech situations. In the SSC-SD, the same speech situations as in the SSC-ER are listed. This time the participant is asked to indicate to which extent they experience speech difficulties in these situations by giving each situation a score on a five-point scale with one corresponding with "no difficulty" and five with "severe difficulty". A score higher than 120.4 on the SSC-SD indicates that the participant has severe difficulties in different speech situations. In the S-scale, 39 communication attitudes are listed, for example "I do not have problems in having a conversations with important people" or "I speak better than I write". Participants were asked to indicate whether this statements are applicable to them (Brutten & Vanryckeghem, 2003). A score over 11.91 indicates severe negative attitudes about speech (Vanryckeghem & Brutten, 2012).

In the BCL, 95 behaviours are listed (for example, "touching your hair" and "pretending not to know the answer" and participants were asked to check whether they used these behaviours to aid them with their speech. The participants then had to score how frequent they used this behaviour on a five-point scale. This questionnaire does not directly investigate emotions about speech, but measures the amount of strategies the participants (consciously) use while speaking. A high score on the BCL indicates more, or more frequent behaviours that aids the speaker with their speech.

Fourteen of the 22 participants that were included in the analysis completed all four questionnaires and five participants completed only three questionnaires. The S-scale was completed by all nineteen participants, the BCL by 16 participants, the SSC-ER and SSC-SD were both completed by 18 participants.

Because all questionnaires investigate roughly the same subject, emotions and attitudes regarding speech, using all questionnaires in one analysis is likely to affect multicollinearity in the statistical analysis. Therefore a correlation matrix between the scores was calculated to investigate which questionnaires to include into the analyses. This revealed that the SSC-ER correlated moderately with the S-scale (.66) and highly with the SSC-SD (.79). The S-scale and the SSC-SD also correlated moderately (.65). The BCL however correlated poorly with all other questionnaires (S-scale: .12; SSC-SD: .22; SSC-ED: .21). As SSC-ER had the highest correlation with S-scale and SSC-SD, and BCL did not correlate with the other questionnaires, it was decided to include both the SSC-ER and the BCL scores in the analyses.

5.7 Statistical analysis

For the statistical analysis of all data, the program R was used (R Core Team, 2013). First, the proportion of NSBs per word was calculated by dividing the total amount of NSBs by the total number of words in the speech sample. The proportion of stutter-like disfluencies and other disfluencies was calculated in the same manner. For every participant a mean severity score and mean duration of NSBs were obtained. The mean durations were log transformed to achieve a normal distribution.

For a subset of the analyses, a composite score of the NSBs was calculated, combining results on the duration of NSBs, severity scores and proportion of NSBs into one value. This follows the same structure used in the SSI-4, where proportion and duration of SLDs together with severity of NSBs are combined into one score (Riley, 2009). In this thesis, however, the proportion of NSBs, mean duration of NSBs and mean severity of NSBs are combined into one score. To make sure all three variables had an equal impact on the combined value, it was decided to multiply these scores rather than adding them like in the SSI. This was done by first transforming the severity score and the duration of NSBs to a score between 0 and 1. The duration of NSBs was divided into four groups based on total range (190 ms – 21089 ms). Durations of the lowest quadrant (190 ms – 5415 ms) were given a score of .25, durations in the second quadrant a .5 (5415 ms - 10640 ms) and so on. The severity score was transformed by dividing the severity score by 4, resulting in a score between 0 - 1, as the highest severity score that was given was 4. The transformed severity scores, duration scores and the proportion of NSBs were then multiplied with each other to calculate the combined value of NSBs. The mean duration of SLDs and ODs in ms was also calculated. All durations were log transformed to achieve normal distribution.

To determine possible predictors of the NSBs across all participants, a backwards stepwise regression analysis was conducted. Possible predictors included in the analyses were the proportion of SLDs, proportion of ODs, age and gender. Multicollinearity was tested by calculating the variance inflation factor, none of the predictors had a variance inflation factor higher than 2.8 indicating low to moderate multicollinearity. The assumptions for the regression analysis were tested by plotting the normal Q-Q, residuals versus fitted, scale location, residuals versus leverage and Cook's distance. These showed that one participant (NS10) could be considered an outlier, possibly because of a high proportion of ODs. As the high proportion of ODs could be considered a natural occurrence in the neurogenic stuttering population, it was decided to not exclude NS10 from the data. For all analyses the influence of outliers differing

over two standard deviation from the mean was investigated by running the analysis a second time without these outliers.

Within the neurogenic stuttering group, a few more possible predictors were investigated with a linear regression: time post-onset, amount of co-occurring speech and language disorders. Although aetiology was hypothesised to be a predictor of NSBs, it was not included in the model as only four participants had an aetiology other than CVA. The linear regression was conducted with the same procedure, however only within the neurogenic stuttering group. As only sixteen participants completed both the SSC-ER and BCL, an analysis with the scores of the BCL and SSC-ER as possible predictors was carried out in a second analysis with a subset of these sixteen participants of the neurogenic stuttering group to investigate the predictive value of emotions and attitudes about stuttering.

5.8 Reliability

Inter- and intrarater reliability data were obtained by rescoring the ODs, SLDs and NSBs of eight randomly selected speech samples (20%; four participants in the control group and four in the neurogenic stuttering group)

For interrater reliability, these speech samples were analysed by a speech language therapist with experience in analysing stuttering behaviour (Vanopdenbosch). To ensure the second rater used the same definitions for all behaviours and severity scores, speech samples of two other videos were analysed together with the first rater. During this practice session, the first and second rater discussed all disagreements until a consensus was made. The interrater reliability was measured by calculating the correlation between the scores of the two raters. The proportion of SLDs of both raters was highly correlated (.89), just as proportion NSB (.92). The proportions of OD had a correlation of .73. The two raters also correlated highly on severity score (.78). The combined NSB score of the two raters also correlated highly (.93). Only mean duration of NSBs correlated very poorly (.01). This indicates that both raters analysed the video's similarly on all measurements except duration of NSBs.

Next to an interraterreliability, an intraraterreliability of the first rater was determined by scoring eight videos for a second time. All measurements of the first and second analysis correlated highly: proportion of SLD: .99, proportion OD: .94, proportion NSB: .95, duration of NSB: .89, severity score of NSB: .81 and the combined score of NSB: .97. This indicates that the rater was highly consistant in analysing the videos.

6. Results

In this section, first the descriptive statistics of the data will be reported to give an overview of the results. Next, results of the regression analyses of possible predictors of non-stuttering behaviours will be described for all participants. After this, the results of the variables that may influence non-speech behaviours in the neurogenic stuttering participants only will be described.

6.1 Descriptive statistics

The descriptive statistics per variable for all participants are displayed in table 5. Because information on co-occurring disorders, time post-onset and the emotions and attitude scores are only available for part of the neurogenic stuttering group, these are displayed separately in table 6.

Table 5 Median, standard deviation and range for the proportion and mean duration of normal disfluencies and stuttering-like disfluencies, and the proportion, mean duration, mean severity score and composite score of the non-speech behaviours of all participants

	Neurogenic stuttering group		**Control group**	
	Median (SD)	**Range**	**Median (SD)**	**Range**
Other disfluencies				
Proportion	.06 (.14)	.01 – .71	.04 (.02)	.01 - .08
Mean duration (ms)	785 (238)	520 – 1331	664 (84)	510 – 857
Stuttering-like disfluencies				
Proportion	.07 (.13)	.02 – .55	.01 (.01)	.00 - .04
Mean duration (ms)	866 (693)	493.67 – 3664	667 (354)	355 - 1737
Non-speech behaviours				
Proportion	.26 (.19)	.07 - .99	.18 (.06)	.09 - .30
Mean duration (ms)	1680 (951)	1044- 5652	1283 (316)	889 – 1996
Mean severity score	1.17 (.25)	1 – 1.99	1.05 (.07)	1.00 - 1.25
Composite score	.02 (.02)	.0028 -.091	.0117 (.0045)	.0054 - .0209

Table 6 Median, standard deviation and range for the co-occurring disorders, time post-onset and emotions and attitude scores for the 22 participants with neurogenic stuttering

	Median (SD)	**Range**	**Cut-off score developmental stuttering**
Co-occurring disorders	1 (.77)	0 - 2	
Time post-onset (days)	76.5 (281)	2 – 1274	
Score emotions and attitude			
SSC-ER **	85.5 (38.74)	51 - 181	125.71
SSC-SD **	101 (45.38)	51 - 182	120.4
S-scale *	19 (7.21)	6 - 35	11.91
BCL ***	18 (35.04)	0 - 151	19.38

* Data only available for 19 neurogenic stuttering participants
** Data only available for 18 neurogenic stuttering participants
*** Data only available for 17 neurogenic stuttering participants

As can be seen in appendix B, four participants (C1, C2, C11 and C19) from the control group had SLD proportions over .03. This means that they have higher SLD frequencies than the cut-off score commonly used as a diagnostic criteria for stuttering, as they have a frequency of SLDs over 3%. As they were not diagnosed with (neurogenic) stuttering nor any other speech- and language disorder prior to recording they are considered natural occurrences within the group of healthy elderly speakers. In contrast, NS6 had a SLD frequency below 3%. This participant was diagnosed with neurogenic stuttering, based on a SLD frequency higher than 3% in a reading task. These issues will be discussed further in chapter 7.

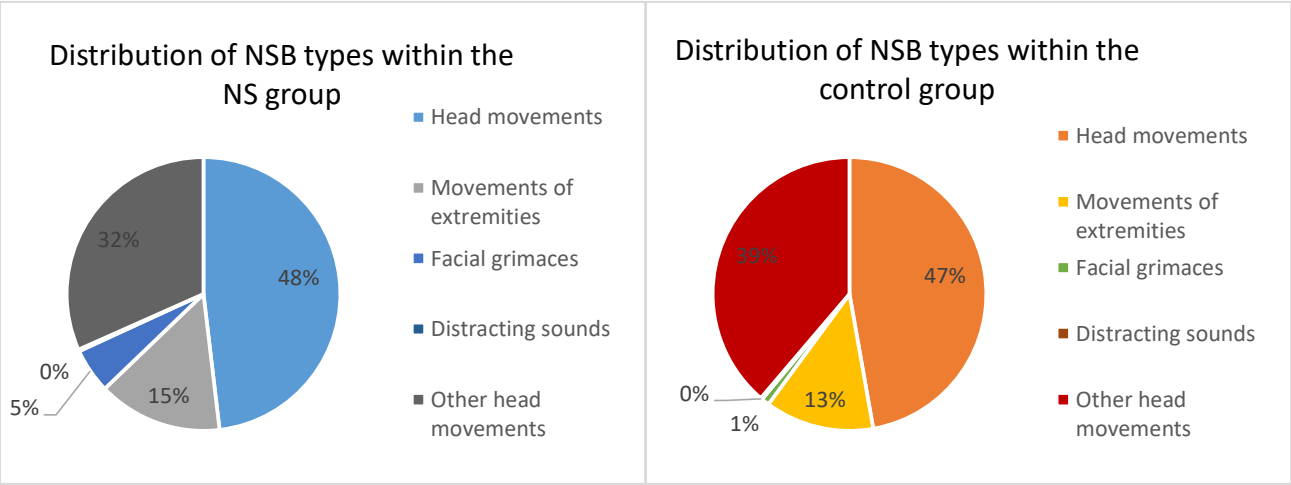

Figure 1, Distribution of NSB types in the neurogenic stuttering group (left) and the control group (right).

In figure 1 the distribution of NSB types per group is displayed. *Head movements* were all movements of the head, such as 'head moving forward' and 'turning away'. *Other head movements* falls under the *head movements* category, but is displayed separately because it was the most annotated non-speech behaviour. *Other head movements* were all head movements that did fit any of the described *head movements* in table 4 (*head moves back, head moves forward, turning away, poor eye contact or constant looking around*). As can be seen in figure 1, the NSB types are distributed similarly in the NS group and the control group, with *head movements* and *other head movements* as the most frequent scored behaviour. *Movement of extremities*, such as *arm movement* and *torso movement*, accounted for respectively 15% and 13% of all non-speech behaviours. *Facial grimaces* (e.g. *grimacing*, *eye blinking*) were seen more often in the neurogenic stuttering group (5% of all NSBs) than in the control group (1% of all NSBs). In both groups, *distracting sounds* (e.g. *whistling*, *sniffing*) were annotated the least, with 0.3% in both the neurogenic stuttering group and the control group.

The results of the neurogenic stuttering and the control group are displayed in several plots. In figure 2, proportion of NSBs, SLDs and ODs are displayed, and in figure 3, the mean duration of these behaviours is shown. The NSB proportion within the control group was lower (M = .18, SD = .06) compared to the neurogenic stuttering group (M = .28, SD = .19). The proportion of SLDs was also lower in the control group (M = .02, SD = .02) compared to the neurogenic stuttering group (M = .12, SD = .13).The proportion of ODs was, again, higher in the neurogenic stuttering group (M = .09, SD = .14) compared to the control group (M = .04, SD = .02). As can be seen in figure 2, the notches of the proportion NSB and proportion SLD do not overlap which is evidence for a significant difference between the medians of both groups. This is also the case for the mean duration of NSBs with a longer duration within the neurogenic stuttering group (M = 1894, SD = 951) compared to the control group (M = 1360.85, SD = 315.51). Duration NSB notches also show no overlap between both groups. The duration of SLDs and ODs was longer within the neurogenic stuttering group (M_{SLD} = 1064, SD_{SLD} = 693; M_{OD} = 850.64, SD_{OD} = 238.14) compared to the control group (M_{SLD} = 719.36, SD_{SLD} = 353.45; M_{OD} = 669.33, SD_{OD} = 83.65). The 95% confidence interval of the medians of the duration of SLDs and

the duration of ODs seem to overlap, as can be seen in figure 3. There are several outliers to be seen in both groups, the influence of these outliers will be addressed later. In addition, all NSBs were given a severity score ranging from one to five. As can be seen in figure 4, the mean severity score is higher in the neurogenic stuttering group compared to the control group, with a wider range.

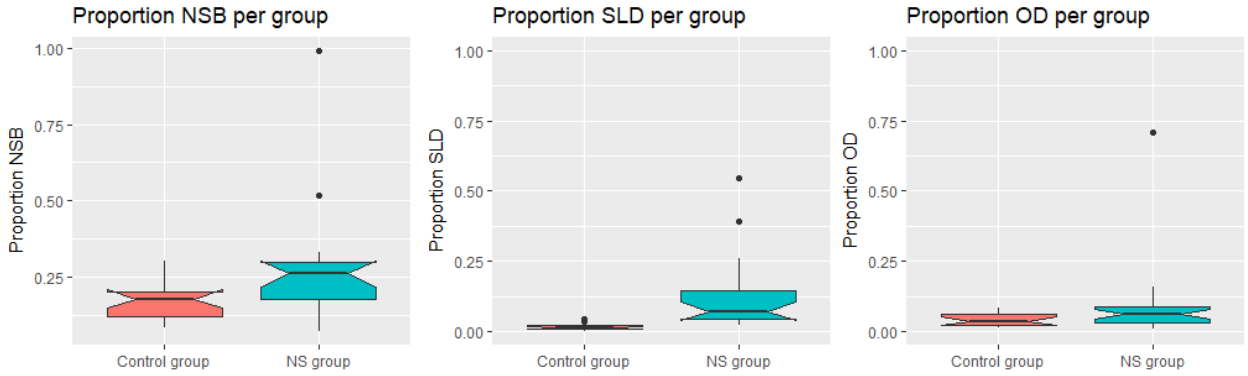

Figure 2, Proportion of non-speech behaviours (NSB) (left), stuttering-like disfluencies (SLD) (middle) and other disfluencies (OD) (right), per group

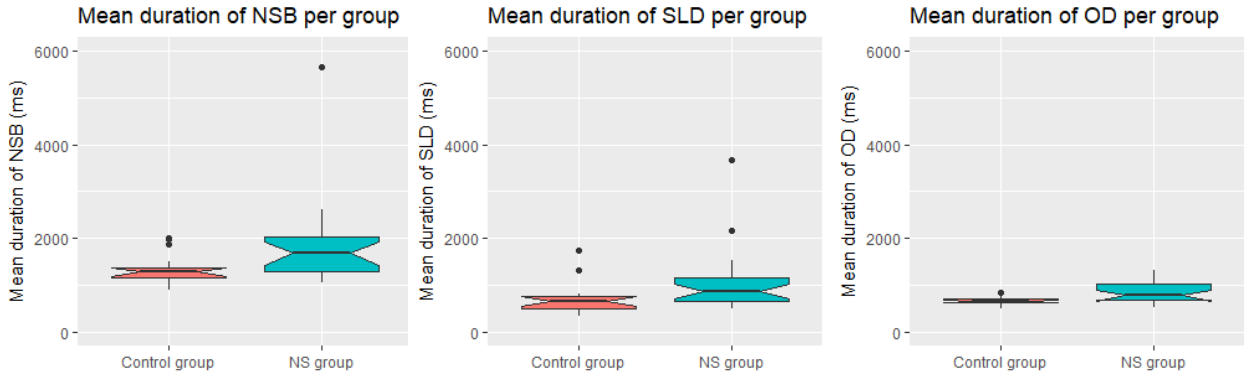

Figure 3, Mean duration of non-speech behaviours (NSB) (left), stuttering-like disfluencies (SLD) (middle) and other disfluencies (OD) (right), per group

A combined NSB score, combining the proportion, duration and severity score, was calculated for all participants. As can be seen in figure 4, the neurogenic stuttering group scored, on average, higher on the combined NSB measure (M = .02, SD =.02) than the control group (M = .0123, SD = .0045). The range of scores in the NS group was wider, as was the case with all other measures.

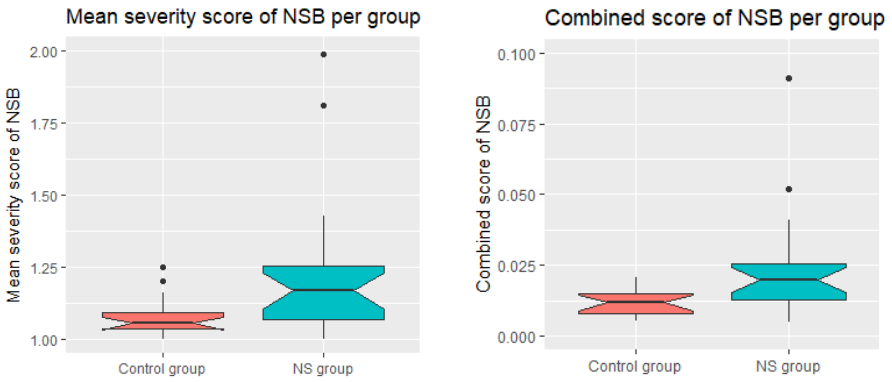

Figure 4, Mean severity score of non-speech behaviours (NSB) per group (left), and the combined value of non-speech behaviours (NSB) per group (right)

The average number of NSBs that were seen during SLDs and ODs are shown in figure 5. This represents a measure of the overlap between NSBs and disfluencies, rather than the occurrence of NSBs over the entire speech sample. The average amount of NSBs per SLD was slightly higher within the neurogenic stuttering group (M = .88, SD = 1.08) compared to the control group (M = .76, SD = .39). The range for the amount of NSBs per SLD within the neurogenic stuttering group was wider (0.05 – 5.50) compared to the range within the control group (.00 – 1.67). The number of NSBs per OD was also slightly higher within the neurogenic stuttering group (M = .86, SD = 1.16) compared to the control group (M = .64, SD = .24). The range for the neurogenic stuttering group was .04 to 5.81 NSBs per OD, which was again wider compared to the range of the control group (.18 – 1.00).

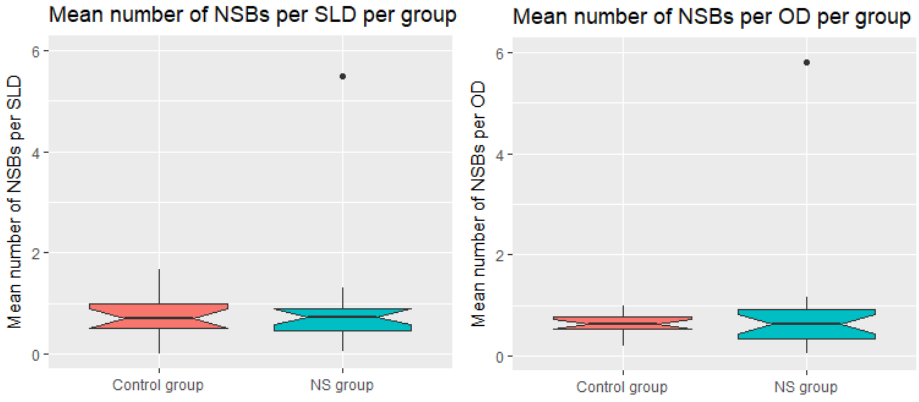

Figure 5, Mean number of NSBs per SLD (left) and ODs (right), per group

6.2 Predictors of non-speech behaviours

This section is divided into two parts. The first part describes the analyses that were done with the data of both the neurogenic stuttering group and control group. These analyses test the relationship between stuttering characteristics and non-speech behaviours of participants with and without neurogenic stuttering. The second part describes the analyses within the neurogenic

stuttering group only. This analysis will additionally focus on the variables specific to the group of stuttering participants, such as the aetiology and time post-onset of the neurological event leading to their stuttering.

6.2.1 Predictors of non-speech behaviours for all participants

To investigate whether the proportion of stuttering-like disfluencies (SLD) would predict non-speech behaviours (NSBs), a backwards regression analysis for each measure of non-speech behaviours (proportion, duration, severity and the combined score) was conducted. Based on previous literature, proportion of other disfluencies (OD), age and gender were also added to the model as other possible predictors. These regression analyses will be described separately in each of the sections below.

6.2.1.1 Proportion of non-speech behaviours

A backwards regression analysis was carried out, with proportion of NSBs as the dependent factor and proportion of SLD, proportion of OD, age and gender as possible predictors. After manual exclusion of non-significant predictors, the final model explained 79% of the variance ($R^2_{adj} = .79$, $F(2, 36) = 71.34$, $p < .01$). There were two significant predictors of the proportion of NSBs: SLD proportion ($\beta = .4$, $t = 3.39$, $p < .01$) and OD proportion ($\beta = .54$, $t = 4.57$, $p < .001$). This indicates that when the proportion of SLD or OD increased, the proportion of NSB increased as well. An ANOVA between models with all predictors and the model with only the significant predictors showed that there was no significant difference between these models ($F(-2, 36) = 0.48$, $p = .62$). This indicates that having age and gender as a predictor in the model does not greatly affect the outcome. The relationship between the NSB proportion and SLD proportion and between NSB proportion and OD proportion are shown in figure 6. In the right-sided plots, the same relationships within the two groups are represented. NS10 and NS4 can be labelled as outliers as their proportion SLD and NSB scores differ more than two standard deviation from the mean. The proportion OD for NS10 also differed more than two standard deviation from the mean. To test for the influence of these outliers on the model, the regression analysis was ran again without these outliers. The analysis excluding both outliers resulted in a model that explained 17% of the variance ($F(1, 35) = 8.4$, $p < .001$), with proportion of OD as the only significant predictor ($\beta = .44$, $t = 2.99$, $p < .001$). Another analysis, excluding only NS10, resulted

in a model with proportion of SLD as a significant predictor of NSB proportion (R_{adj} = *.37*, F_{model}(*1,36*) = *22.69*, p_{model} <*.001* ; β_{SLD} = *.62*, t_{SLD} = *4.76*, p_{SLD} <*.001*).

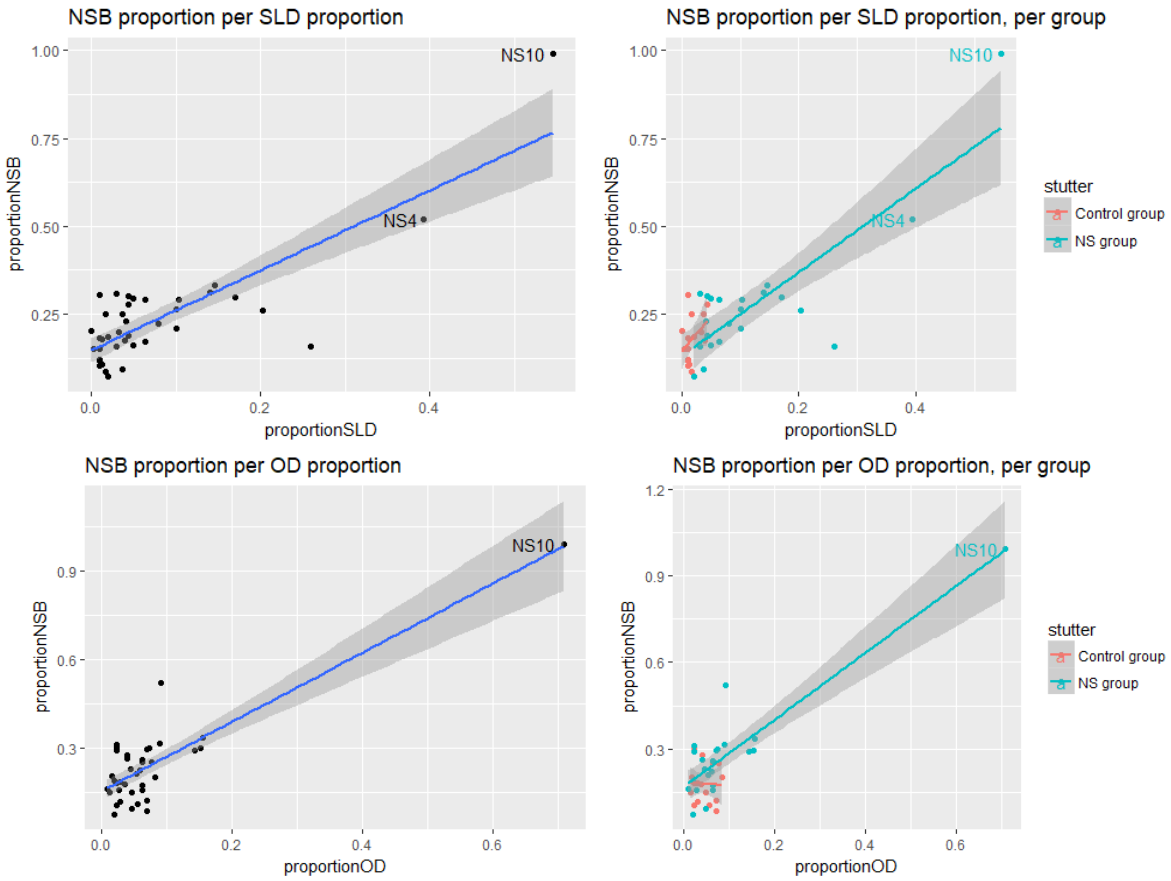

Figure 6, plots of the relationship of the proportion of stuttering-like disfluencies (SLD) (above) and other disfluencies (OD)(below) with the proportion of non-speech behaviours (NSB). Right-sided plots show this relationship within the neurogenic stuttering group (blue) and the control group (red).

6.1.2.2 Duration of non-speech behaviours

As with proportion of non-speech behaviours, a backwards regression analysis was carried out with duration of NSBs as dependent variable and proportion of SLD, proportion of OD, age and gender as possible predictors. Backward regression resulted in a model (R^2_{adj} = *.16*, F(*1, 37*) = *8.41*, p < *.01*) with SLD proportion as the only significant predictor of the duration of NSB (β =*.43*, t = *2.9*, p < *.01*). As can be seen in figure 7, an increase in the SLD proportion resulted in longer durations of NSBs. An ANOVA between the models including all predictors and the final model was not significant (F(*-3, 37*) = *1.7*, p = *0.18*), showing that the predictors other than SLD did not affect the model.

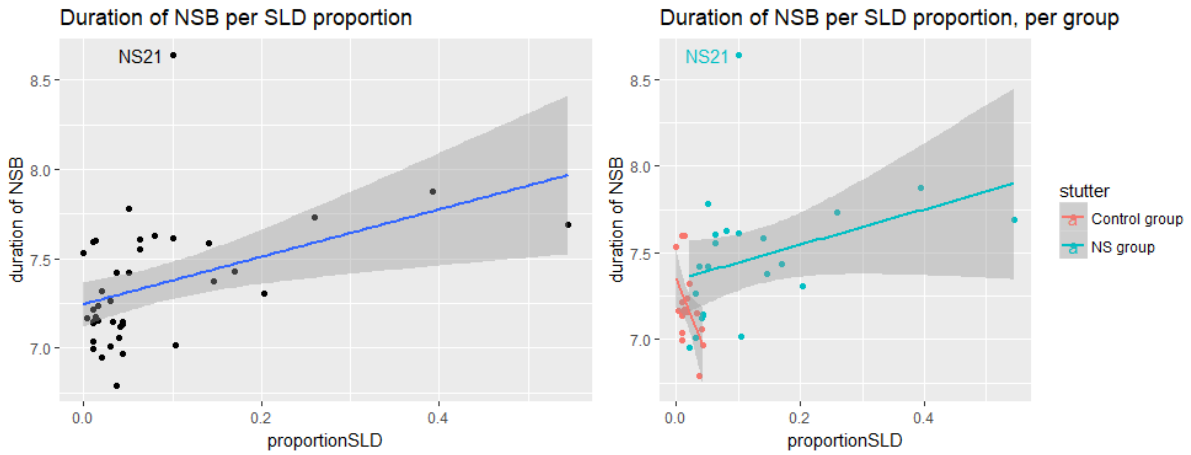

Figure 7, plots of the relationship between the duration of non-speech behaviours (NSB) and the proportion of stuttering-like disfluencies (SLD). Right plot shows this relationship within the neurogenic stuttering group (blue) and the control group (red).

One participant, NS21, had a mean duration that differed more than two standard deviation from the mean, and could therefore be identified as an outlier. To test the influence of this outlier, the analyses was ran again without NS21. This resulted in a final model (R_{adj} = *.33*, F(*2, 35*) = *10.16*, p < *.001*) with both proportion of SLD and gender as significant predictors (β_{SLD} = *.54*, t_{SLD} = *4.03*, p_{SLD} <.001 ; β_{male} = *-.3*, t_{male} = *-2.28*, p_{male} = *.03*). This suggests an effect of gender where female participants displayed NSBs with longer durations than male participants. As the adjusted r-squared had the highest value in the model without the outliers, this might be the best fitting model.

6.1.2.3 Severity of non-speech behaviours

Backwards regression was performed with the severity score of the NSBs as dependent factor and the same predictive variables (proportion SLD, proportion OD, age and gender). The only significant predictor of the severity score was age (β = *-.48*, t = *-3.3*, p < *.01*), with an increase of age resulted in a decrease in severity scores. The model explained 20% of the variance (R^2_{adj} = *.20*, F(*1, 37*) = *11*, p < *.01*). The relationship between severity score and age is pictured in figure 8. In a model with both proportion SLD and age (R^2_{adj} = *.26*, F(*1, 36*) = *7.61*, p < *.01*), the proportion of SLDs nearly achieved significance (t = *1.86*, p = *.07*). This trend can be seen in figure 9. An ANOVA between the models showed an almost significant difference between the models (F(*1, 36*) = *3.48*, p = *0.07*). An ANOVA between the model with age and proportion SLD as predictor and the first model with four predictors was highly non-significant (F(*2,34*) = *.048*, p = *.95*). This indicates that there is a big difference between these models, and the model with both age and proportion SLD might be a better fit.

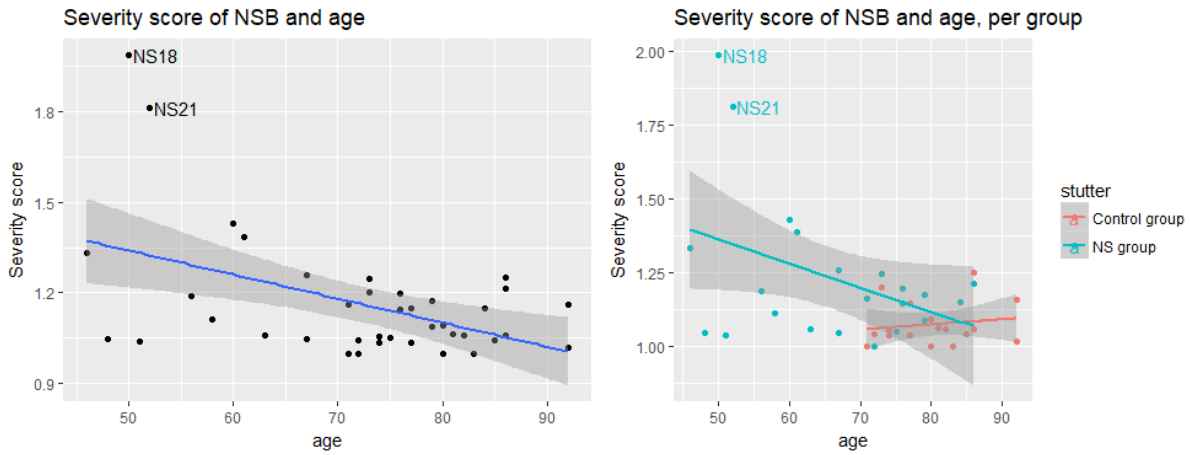

Figure 8, plots of the relationship between severity score of non-speech behaviours (NSB) and age. Right plot shows this relationship within the neurogenic stuttering group (blue) and the control group (red).

Figure 9, plot of the (non-significant) relationship between the severity score of non-speech behaviours (NSB) and the proportion of stuttering-like disfluencies (SLD).The right plot shows this relationship within the neurogenic stuttering group (blue) and the control group (red).

There are two participants, NS18 and NS21, which could be considered outliers as they vary more than two standard deviation from the mean severity score. While the scores of these participants can be seen as natural variances these are natural variances that occur within the population, the analysis was rerun without NS18 and NS21 to test for the influence of these outliers on the analysis. This resulted in a final model (R_{adj} = *.45*, F(*1*, *35*) = *29.87*, p < *.001*) with proportion SLD as the only significant predictor (β = *.67*, t = *5.46*, p <*.001*). This indicates that these outliers did have influence on the analyses and as the adjusted R-square was highest in the model without the outliers, this model might be the best fit out of all models with severity score as the dependent factor.

6.1.2.4 Combined score of non-speech behaviours

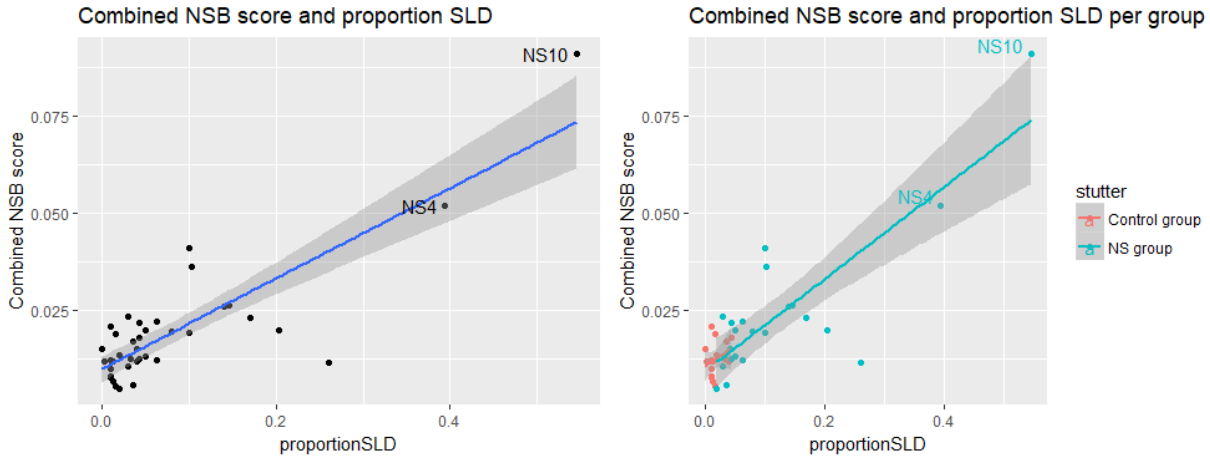

Figure 10, plots of the relationship between the combined score of non-speech behaviours and the proportion of stuttering-like disfluencies (SLD). Right plot shows this relationship within the neurogenic stuttering group (blue) and the control group (red).

As described in the methods section, the proportion, duration and severity of NSBs were combined into one composite value per participant. A backwards regression analysis was executed in the same way as previously done with all NSB components separately: the combined score as dependent value and proportion of SLD, proportion of OD, gender and age as possible predictors. The backwards regression resulted in a model that explained 77% of the variance ($F(2, 36) = 66.23$, $p < .001$). The SLD proportion was a significant predictor of the combined NSB score ($\beta = .5$, $t = 4.05$, $p < .001$), as was the OD proportion ($\beta = .45$, $t = 3.64$, $p < .001$). Higher proportions of SLD and OD resulted in an increase in the combined value of NSB. These relationships are presented in figure 10 and figure 11. An ANOVA comparing the model including all predictors and the final model was not significant ($F(-2, 36) = 2.1$, $p = .14$).

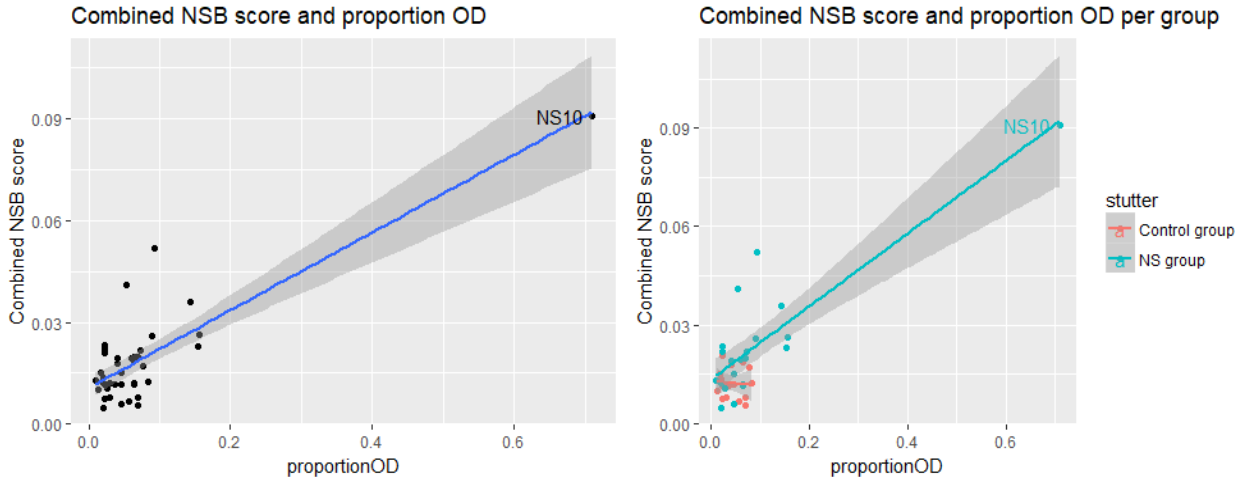

Figure 11, plots of the relationship between the combined score of non-speech behaviours (NSB) and the proportion of other disfluencies (OD). Right plot shows this relationship within the neurogenic stuttering group (blue) and the control group (red).

Participant NS10 could be identified as an outlier as the combined NSB score differed more than two standard deviation from the mean. An analyses without this participant resulted in

a model that explained 47% of the variance ($F(2, 35) = 18.03$, $p < .001$). In this model, both proportion SLD and age were significant predictors of the combined NSB score ($\beta_{SLD} = .59$, $t_{SLD} = 4.73$, $p_{SLD} < .001$; $\beta_{age} = -.26$, $t_{age} = -2.12$, $p_{age} = .04$). Again, an increase in SLD resulted in an increase of the combined value of NSB, but an increase in age resulted in a decrease of the combined NSB value.

6.2.2 Predictors of non-speech behaviours within the neurogenic stuttering group

To further investigate the possible predictors of non-speech behaviours (NSB) within people with neurogenic stuttering, two analyses were done with data of the stuttering group only. The possible predictors were variables that were only relevant for the participants with neurogenic stuttering: time post stuttering onset, number of co-occurring speech and/or language disorders and emotions and attitudes related to their speech. The first analysis was with all participants of the neurogenic stuttering group, with time post stuttering onset, number of co-occurring speech and/or language disorders, proportion of stuttering-like disfluencies (SLD), proportion of other disfluencies (OD), age and gender as variables. The second analysis was within a subset of the neurogenic stuttering group, and only included those who had completed the questionnaires about emotions and attitudes. To analyse the influence of the emotions and attitudes related to the stuttering, a backwards regression was carried out including the results of the questionnaires as a variable. As the results in 6.2.1 showed that duration and severity of NSBs did not have a high predictive value within the total group of stuttering and non-stuttering participants, proportion of NSBs and the combined NSB value were used as the dependent variable in these analyses.

6.2.2.1 Predictors of the proportion of non-speech behaviours in all neurogenic stuttering participants

Backwards regression with proportion SLD, proportion of OD, gender, age, time post-onset, number of co-occurring speech and/or language disorders resulted in a model that explained 83% of the variance ($F(2, 19) = 53.35$, $p < .001$). The proportion of SLD was a significant predictor of the proportion of NSBs within the neurogenic stuttering group ($\beta = .34$, $t = 2.32$, $p = .03$) as was proportion of OD ($\beta = .63$, $t = 4.38$, $p < .001$). As can be seen in figure 12, an increase in proportions of SLD and OD resulted in an increase of the NSB proportion. An ANOVA between the first and final model of the backwards regression showed no significant difference between the models ($F(-7, 19) = 1.56$, $p = .24$).

To test for the influence of outliers, the analyses was run again without outliers. Removing both NS10 and NS04 from the analyses, resulted in a model with only proportion OD as a significant predictor ($R^2_{adj} = .26$, $F_{model}(1,18) = 7.8$, $p_{model} = .01$; $\beta_{OD} = .55$, $t_{OD} = 2.8$, $p_{OD} = .01$). An analyses where only NS10 was removed from the dataset resulted in a model that explained 44% of the variance ($F(3, 17) = 6.28$, $p < .01$). In this model, proportion OD was a predictor of the proportion of NSB ($\beta = .61$, $t = 3.58$, $p < .01$). Both gender and the number of co-occurring speech and language disorders were also significant predictors, with females having a higher proportion of NSB than males and an increase in the amount of co-occurring disorders resulting in an increase of the NSB proportion ($\beta_{male} = -.38$, $t_{male} = -2.2$, $p_{male} = .045$; $\beta_{co\text{-}occurring} = 0.5$, $t_{co\text{-}occurring} = 2.79$, $p_{co\text{-}occurring} = .01$).

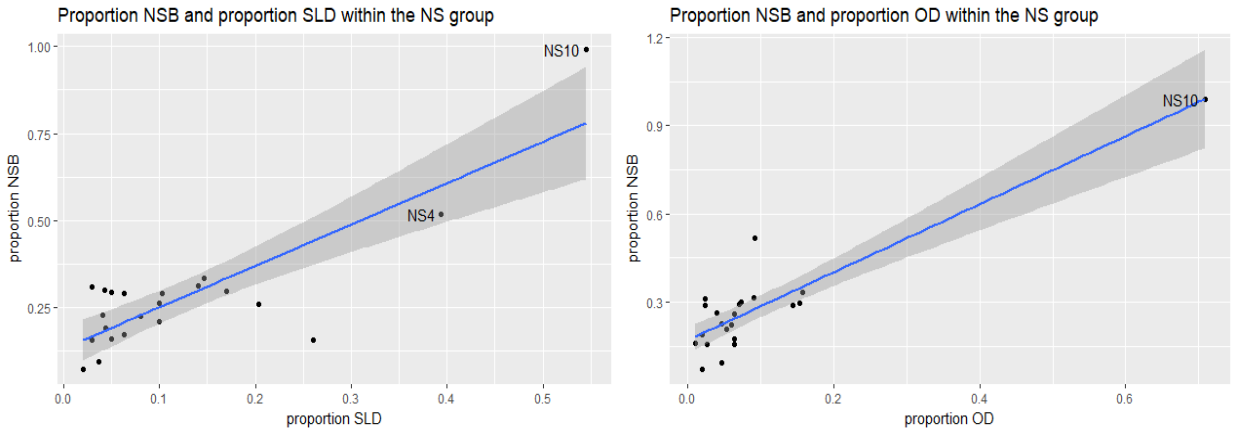

Figure 12, the relationship between the proportion of non-speech behaviours and the proportion of stuttering-like disfluencies (SLD) (left), and proportion of non-speech behaviours with other disfluencies (OD) (right), within the neurogenic stuttering group

6.2.2.2 Predictors of the combined value of non-speech behaviours in all neurogenic stuttering participants

Another backwards regression with proportion SLD, proportion of OD, gender, age, time post-onset and number of co-occurring speech and/or language disorders as possible predictive values was carried out with the combined NSB value as dependent factor. This resulted in a model that explained 77% of the variance ($F(2, 19) = 36.35$, $p < .001$). The proportion of SLD was a significant predictor of the combined NSB value within the neurogenic stuttering group ($\beta = .41$, $t = 2.4$, $p = 0.26$) as was proportion of OD ($\beta = .53$, $t = 3.2$, $p < .01$). Similar to the results in section 6.2.1.4 an increase in SLD or OD proportions resulted in an increase in the combined NSB value, both relationships are shown in figure 13. An ANOVA between the first (including all possible predictors) and final model of the backwards regression showed no significant difference between the models ($F(-4, 19) = .79$, $p = .54$).

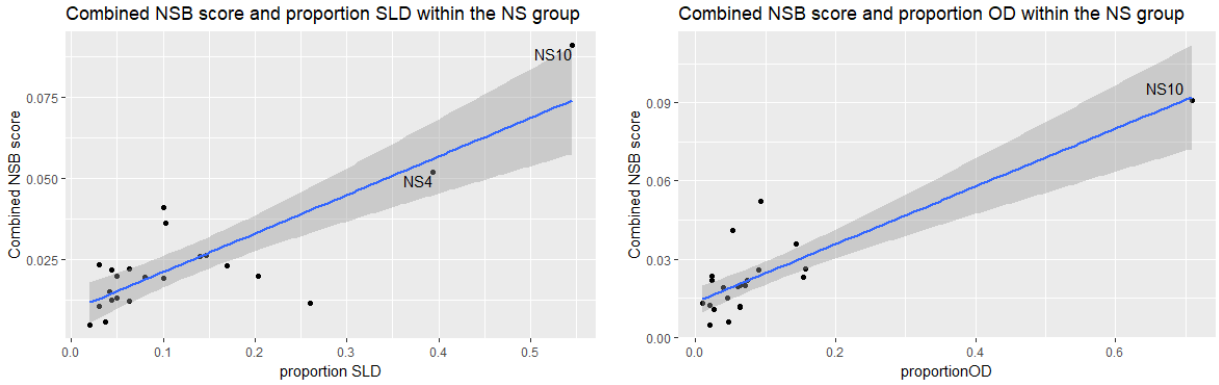

Figure 13, the relationship between the combined NSB score and the proportion of stuttering-like disfluencies (SLD) (left), and proportion of non-speech behaviours with other disfluencies (OD) (right), within the neurogenic stuttering group

To test for the influence of outliers, the analysis was run again without NS10. This resulted in a final model with proportion of SLD a significant predictor of the combined NSB value ($R_{adj} = .32$, $F_{model}(1,19) = 10.56$, $p_{model} < .01$; $\beta_{SLD} = .60$, $t_{SLD} = 3.25$, $p_{SLD} < .01$).

6.2.2.3 Relationship between scores of the emotions and attitude questionnaires and non-speech behaviours

A backwards regression was carried out with a subset of 16 participants of the neurogenic stuttering group. These participants had completed both the Behavioural Checklist (BCL) and the Speech Situation Checklist – Emotional Reactions (SSC-ER) questionnaires. The BCL and SSC-ER scores were included as possible predictors of the proportion of NSBs, in addition to proportion SLD, proportion OD, gender, age, time post-onset and number of co-occurring speech and/or language disorders.

The first backwards regression was run with proportion of NSB as the dependent factor. The final model explained 89% of all variance ($F(2, 13) = 65.49$, $p < .001$). Similarly to the previous models, the final model only had proportion SLD and proportion OD as significant predictors of proportion NSB ($\beta_{SLD} = .42$, $t_{SLD} = 3.01$ $p_{SLD} < .01$; $\beta_{OD} = .59$, $t_{OD} = 4.34$, $p_{OD} < .001$). This means that higher SLD proportions and OD proportions resulted in higher proportions of NSB. An ANOVA including the final model and the model including all variables showed that the variables other than proportion of SLD and proportion of OD did not contribute to the model ($F(-6, 13) = 1.72$, $p = .25$), indicating that the model with only proportion SLD and proportion OD as predictors of proportion NSB was the best fitting model.

This model was rerun to test for the influence of outliers. First only NS10 was removed from the analysis, resulting in a model with only proportion SLD as a significant predictor of the NSB proportion ($R^2_{adj} = .5$, $F(1, 13) = 14.92$, $p < .01$; $\beta_{SLD} = .73$, $t_{SLD} = 3.86$, $p_{SLD} < .01$). An ANOVA between the model including all variables and the model with only proportion SLD was not significant, indicating that all the other variables did not contribute to the model ($F(-7, 13) = 1.83$, $p = .24$). However, the ANOVA between the final model and the model including proportion OD almost reached significance ($F(-1, 13) = 4.09$, $p = .09$). The model without both NS10 and NS4 resulted in a model with only proportion of OD as a significant predictor of the NSB proportion ($R^2_{adj} = .5$, $F(1, 12) = 13.52$, $p < .01$; $\beta_{OD} = .73$, $t_{OD} = 3.67$, $p_{SLD} < .01$). Again, an ANOVA between the model including all variables and the final model did not reach significance ($F(-7, 12) = .95$, $p = .54$).

Another backwards regression was run with the combined NSB value as the dependent factor. This issued similar results to the model with proportion of NSB. The final model explained 80% of all variance ($F(2, 13) = 31.27$, $p < .001$). Again, both SLD proportion and OD proportion significantly predicted the combined NSB value, with higher disfluency proportions resulting in a higher combined NSB value ($\beta_{SLD} = .48$, $t_{SLD} = 2.6$ $p_{SLD} = .02$; $\beta_{OD} = .47$, $t_{OD} = 2.54$, $p_{OD} = .02$). An ANOVA between the final model and the model including all variables was highly insignificant ($F(-6, 13) = .37$, $p = .87$). This regression was also rerun without NS10 to test for the influence of this outlier, resulting in a model with only proportion SLD as a significant predictor of the combined NSB value ($R^2_{adj} = .40$, $F_{model}(1, 13) = 10.56$, $p_{model} < .01$; $\beta_{SLD} = .66$, $t_{SLD} = 3.25$, $p_{SLD} < .01$).

7. Discussion

This study was carried out to determine whether people with neurogenic stuttering display non-speech behaviours, like grimacing or head movements, during speech and what factors predict these non-speech behaviours. Conversational speech of 22 people with neurogenic stuttering and 17 elderly healthy control speakers was analysed on stuttering-like disfluencies (SLD), other disfluencies (OD) and non-speech behaviours (NSB). The results show that participants with neurogenic stuttering do indeed show more NSBs on average than participants in the control group, contradicting the widely quoted characteristics proposed by Canter (1971) and Helm-Estabrooks (1999). This indicates that it is important for both researchers and speech- and language therapist working with neurogenic stuttering patients to address non-speech behaviours. The results will be further discussed in the following paragraphs, followed by a discussion on the qualities and limitations of this study. Finally, the most important results and discussions will be summarised in the conclusion.

7.1 Characteristics of neurogenic stuttering

Developmental stuttering, a stuttering disorder with an onset during childhood, is characterised by SLDs during speech. The presence of non-speech behaviours, such as eye blinking and grimacing, and anxiety about speech in people with developmental stuttering is widely accepted (Logan, 2015; Yairi & Seery, 2015). Often, non-speech behaviours are seen as learned behaviours initially used to help get out of stutters (Yairi & Seery, 2015). However, non-speech behaviours can be present at onset of developmental stuttering, suggesting that non-speech behaviours are an intrinsic symptom of stuttering (Conture & Kelly, 1991; Logan, 2015; Schwartz et al., 1990; Yairi & Ambrose, 2005; Yairi & Seery, 2015).

It is often reported that the characteristics of neurogenic stuttering differ from the characteristics of developmental stuttering. The list of characteristics by Canter (1971) and later revised by Helm-Estabrooks (1999) stated that both non-speech behaviours and anxiety are not present in people with neurogenic stuttering. However, correctly identifying developmental stuttering or neurogenic stuttering based on observation alone proved to be difficult for speech-and language pathologists, suggesting that characteristics of both stuttering disorders are similar (Van Borsel & Taillieu, 2001). Almost all information available on neurogenic stuttering was based on case-studies. Of the 86 cases described in 60 articles, presence or absence of non-speech behaviours were reported in 47 cases and emotions and attitudes in only 21 cases. Non-speech behaviours were present in 40% of the cases that explicitly reported on them and 61% of the cases had an emotional reaction about their speech. One survey study by Theys et al. (2008) reported the presence of non-speech behaviours in 55% of the reported cases.

This is the first study systematically studying non-speech behaviours in people with neurogenic stuttering. As can be seen in appendix B, eleven neurogenic participants in this study (58%) could be identified as presenting with NSBs based on high proportion of NSBs and/or high severity scores (NS10, NS11, NS13, NS16, NS17, NS18, NS19, NS2, NS21, NS4 and NS5). Three participants (NS8, NS9 and NS22) had very high mean durations of NSBs but not high

NSB proportions or severity scores. However, as duration had a very low interrater reliability, it may not be reliable to base occurrence of NSBs based on duration measurements alone. This means 58% of the neurogenic stuttering participants in this study presented with NSBs. This is similar to the study by Theys et al. (2008) where 55% of the reported cases presented with NSBs. In contrast, NSBs occurred in only 40% of the case-studies that explicitly reported NSBs in people with neurogenic stuttering. However, as 47% of the available case-studies do not report on NSBs, it is possible that the actual number of cases described in articles that present with NSBs is much higher, but that the researchers did not report them. As the characteristics by Helm-Estabrooks (1999) are still widely accepted, researchers might not expect NSBs in people with neurogenic stuttering and therefore not notice or report them.

The presence of anxiety about speech and the social consequences of stuttering in developmental stuttering is widely accepted. This is not the case, however, for neurogenic stuttering, where one of the often cited characteristics states that people with neurogenic stuttering do not appear anxious (Helm-Estabrooks, 1999). In the literature, however, almost half of the cases that report emotions and attitudes about stuttering, displayed emotional reactions to their stuttering. The severity of these reactions varied from mild annoyance (Koller, 1983) to anger and hatred about the way of speaking (Stewart & Grantham, 1993). Similar to the findings in the literature, the emotions and attitudes about stuttering varied widely in the neurogenic stuttering group in this study, as can be seen in appendix B. While nine participants seemed to have no or few emotional reactions to their stuttering, four scored above the cut-off score on the SSC-ER indicating severe emotional reactions to their stuttering (NS16, NS21, NS22, NS3). Another five scored between one and two standard deviations higher than the mean of healthy speakers, indicating a mild emotional reaction (NS10, NS20, NS7, NS8, NS9). This is consistent with the literature as half of the neurogenic stuttering participants had mild to severe emotional reactions to their stuttering. All participants that completed the S-scale questionnaire scored above the cut-off score, except two (NS13 and NS5). Six participants who did not score above the cut-off score on the SSC-ER did score above the cut-off score on the SSC-SD (NS14, NS18, NS20, NS9, NS10, NS7). This gives evidence contradicting the characteristic of Helm-Estabrooks (1999), that people with neurogenic stuttering can be anxious about their stuttering.

In developmental stuttering, trait anxiety is found in all levels of stuttering severity, but state anxiety in social communication is higher among persons with severe stuttering compared to persons with mild stuttering (Ezrati-Vinacour & Levin, 2004). Interestingly, amongst the neurogenic stuttering participants, high scores on the emotions and attitudes questionnaires did not always correspond with high NSB measurements. Both NS3 and NS7 both had high scores on at least one of the emotions and attitudes questionnaires, but did not present with high NSB proportions, high severity scores or long durations. They also had relatively low proportions of SLD, respectively .041 and .037. This indicates that although all of these objective measurements indicate a mild neurogenic stuttering disorder, the emotions and attitudes associated with speech of someone with neurogenic stuttering can be severe. In contrast, high proportions of SLD or NSB, high severity rates or duration of NSBs did not immediately result in high scores on the emotions and attitudes questionnaires. NS4, for example, had a high SLD proportion of .39 and also scored high on all three NSB measurements. In her case, this did not result in severe

emotions and attitudes about her speech as she scored relatively low on all three questionnaires that she completed. The same is true for NS17 and NS11. This indicates that emotions and attitudes about speech can be present in people with neurogenic stuttering, even though the stuttering might seem mild. This is especially important for speech- and language therapists working with people with neurogenic stuttering, as it shows that it is essential to identify and address emotions and attitudes of people with neurogenic stuttering about their speech.

There is evidence that social anxiety in developmental stuttering develops as a result from stuttering (Alm, 2014). If this is also the case for neurogenic stuttering, time post-onset might be related to anxiety levels. Of the participants with time post-onsets over six months, two scored above the cut-off score on the SSC-ER (NS21 and NS22), the others also scored above the cut-off score on the S-scale. Interestingly, none of the six participants with time post-onsets under two weeks scored above the cut-off score on the SSC-ER. The possible development of anxiety over time should be addressed in future research but also by speech- and language therapists working with people with neurogenic stuttering.

In short, it is advisable to alter the widely cited characteristics by Helm-Estabrooks (1999) into a list of characteristics that includes non-speech behaviours and anxiety as a possible (but not necessary) characteristics of neurogenic stuttering.

7.1 Predictors of NSBs in all participants

In this study, the predictive value of age, gender, proportion of ODs and SLDs was investigated. As predicted, the proportion of SLDs was a significant predictor of proportion of NSB, meaning that participants with a higher SLD frequency also presented with more NSBs. Interestingly, proportion of OD was also a significant predictor of proportion NSB, with a higher proportion of OD resulting in a higher NSB proportion. This model with proportion of SLD and OD as predictors was a good fit as it explained 79% of the variance. When accepting that people with neurogenic stuttering do present with NSBs, this result is not surprising. NSBs can be linked to moments of disfluency, although they also occur during seemingly fluent moments. More moments of disfluency would then logically result in a higher amount of NSBs. Interestingly, there is no significant difference in amount of NSBs per SLD between the control group, who on average displayed .76 NSB per SLD and .64 NSB per OD, and the neurogenic stuttering group, who displayed .88 NSB per SLD and .86 NSB per OD. This indicates that the control group also displayed on average more NSBs during disfluencies than during fluent speech, as their mean overall NSB proportion was much lower. NSBs might therefore be intrinsic to moments of disfluencies, even in healthy speakers, rather than to stuttering disorders as is proposed by Yairi and Seery (2015) and Logan (2015). These averages in the neurogenic stuttering group are much lower compared to frequency of NSBs per SLD in developmental stuttering, as research on the amount of NSBs in children with developmental stuttering report means ranging from 1.48 NSB per SLD up to 3.18 NSB per SLD (Conture & Kelly, 1991; Yairi et al., 1993). Future research might investigate this possible difference between developmental stuttering and neurogenic stuttering.

The duration of NSB was also significantly predicted by proportion of SLD, indicating that the NSBs were longer in duration in the participants with more SLDs. Removing outliers resulted in a model with gender as a significant predictor. Females had NSBs that were longer in duration compared to males. Severity of NSBs were significantly predicted by age, with younger participants presenting with more severe NSBs. As can be seen in figure 8, this relationship seems not to be present in the control group. This indicates that younger neurogenic stuttering participants displayed more distracting movements compared to the older neurogenic stuttering participants. Proportion of SLDs was almost significant, indicating a trend where a higher proportion of SLD results in more severe NSBs. Removing the outliers, however, resulted in a model with only proportion of SLD as a significant predictor of mean severity of NSBs. Both outliers were relatively young participants, which might explain why age was a significant predictor in the model including both outliers. These results show that participants with a higher frequency of SLDs not only present with more NSBs, but also with longer and more severe NSBs. It is unclear whether this is also seen in people with developmental stuttering, as there is surprisingly little research investigating non-speech behaviours.

Because all movements during speech were analysed, a lot of the analysed movements were movements that normally occur during speech. Therefore, most movements were given a severity score of 1, resulting in low mean severity scores. This, however, does not mean that the participants with neurogenic stuttering did not display distracting or tense non-speech behaviours. As can be seen in appendix B, there were ten neurogenic stuttering participants (45%) and two participants of the control group (12%) with at least one movement that scored a 3 or 4 on the severity scale. However, nine neurogenic stuttering participants (41%) and fourteen participants (82%) of the control group did mostly display non-distracting non-speech behaviours with a severity score of 1. High NSB proportions did not always result in high severity rates, NS20 for example had a relatively high NSB proportion of .293, but only had four movements with a severity score of 2 and all other 63 movements had a severity score of 1. The duration of his movements also weren't long. This might mean that people with neurogenic stuttering sometimes display non-speech behaviours that are associated with their stuttering but are not distracting, tense or long in duration. However, it is also possible that these participants always used a lot of movements during speech and that these movements were not related to stuttering.

The results of the combined NSB score, with proportion, duration and severity of NSB in combined into one value, greatly resemble the results with proportion of NSB as dependent factor. The models with proportion NSB and the combined value also had much higher adjusted R^2, indicating that these models explained more of the variance. The severity and duration measures of non-speech behaviours were time consuming and the interrater reliability of duration was very low. Future studies investigating NSBs might therefore only include proportion and severity of NSBs. However, adding the duration measure does give extra information on characteristics of non-speech behaviours and neurogenic stuttering as three participants had high duration of NSBs but did not have high severity scores or proportions of NSBs. This is the first study on non-speech behaviours that had these separate measurements of NSBs. Studies on NSBs

in developmental stuttering only report the amount of NSBs per disfluency (Conture & Kelly, 1991; Yairi et al., 1993), or give an overall severity score on NSBs rather than a severity score per NSB as in this study (Riley, 2009). It would be interesting to do a similar study on NSBs, measuring proportion, duration and severity of NSBs in children and adults with developmental stuttering.

7.2 Predictors of NSBs within the neurogenic stuttering group

The second aim of this study was to further examine the possible predictors of non-speech behaviours within the neurogenic stuttering group. The hypothesis was that, next to proportion of SLD and OD, time post-onset and emotions and attitudes about their stuttering could be predictors of non-speech behaviours, as one theory about non-speech behaviours is that they develop over time as a reaction to the disfluencies (Guitar, 1998). Other hypothesised predictors were the aetiology and number of co-occurring speech and/or language disorders. Another two backwards regression analyses were carried out with data of only the neurogenic stuttering group. As the questionnaires were not completed by all participants, an analysis was carried out without the questionnaires as to include all neurogenic stuttering participants. Surprisingly, the results of both analyses were similar to the analyses including the control group. Both proportion of SLD and proportion of OD were significant predictors. Excluding the outlier NS10 resulted in a model where next to proportion of SLD, gender and the number of co-occurring disorders were significant predictors of the proportion of NSB. This indicates that females, and participants with more co-occurring disorders had higher proportions of NSBs. Excluding NS4 as well as NS10, however, resulted in a model with only proportion of OD as a significant predictor of NSB proportion. NS4 was both female and had two co-occurring non-speech behaviours, as well as a high proportion of SLDs and NSBs. This might explain why excluding NS10 but not NS4 resulted in a model where gender and number of co-occurring disorders were significant predictors. Further research is needed to fully investigate the influence of co-occurring disorders and gender.

Time post-onset was not a significant predictor of NSBs. This is surprising as a commonly cited theory about non-speech behaviours is that they develop over time (Guitar, 1998). Logically, a longer time post-onset would then result in higher proportions of NSBs or more severe NSBs. This relationship was not found in this study. In children with developmental stuttering, non-speech behaviours can occur right at stuttering onset (Conture & Kelly, 1991; Schwartz et al., 1990; Yairi & Ambrose, 2005). As can be seen in appendix B, of the five participants with a time post-onset under 10 days, three (NS1, NS11 and NS4) displayed severe non-speech behaviours and two of them (NS11 and NS4) had high NSB proportions. Of the six participants with a time post-onset over six months, one (NS18) displayed severe non-speech behaviours and one (NS22) had a relatively high NSB proportion. This shows that in neurogenic stuttering, similarly to in developmental stuttering, non-speech behaviours can be present at stuttering onset. The study of Yairi et al. (1993) showed that the amount of non-speech behaviours per disfluencies in children with developmental stuttering had declined instead of increased in three-month and six-month follow-ups. Theys et al. (2009) found similar results in

their case-study, where they reported a decrease in NSBs in the 3rd test moment compared to the first test moment. However, in three other cases, the amount of NSBs increased over time (Lebrun et al., 1983; Stewart & Grantham, 1993; Vanhoutte et al., 2014). A study with neurogenic stuttering participants including longitudinal analysis of participants with persistent neurogenic stuttering would be very interesting to fully investigate the influence of time post-onset.

Even though aetiology was not included in the analysis, it might be that different aetiologies result in different neurogenic stuttering characteristics. In the study by Theys et al. (2008), participants were divided into four groups based on aetiology. They found that some speech characteristics were more common in some aetiologies compared to others. Single syllable word repetitions, for example, were seen in people with neurodegenerative disease but not in the other aetiology groups and blocks were characteristic for stroke and TBI patients but not for the other two aetiology groups. It is plausible, that when the speech characteristics of neurogenic stuttering are different, the non-speech characteristics could differ as well. Of the studies in the literature that reported the presence of NSBs, 11 cases suffered a stroke, 2 had traumatic brain damage, 2 cases of PD and four cases had other aetiologies such as epilepsy or rotavirus. Of the cases that reported no non-speech behaviours, 34 in total, neurodegenerative diseases were a more frequently aetiology (9 cases). Six cases had traumatic brain injury and stroke was, again, a common aetiology (8 cases). Other reported aetiologies included meningitis and dementia. Although most of these cases are reported in separate case-studies, it gives some evidence that non-speech behaviours might be more common in certain aetiologies like stroke. Characteristics of some aetiologies, for example Parkinson's Disease (PD), could result in less non-speech behaviours. As PD is often characterised by kinematic deficits and rigidity, non-speech behaviours might occur less. In this thesis, out of a total of 22 participants with neurogenic stuttering, 18 participants had suffered a stroke, two had traumatic brain injury, one participant had ALS and one had PD. Strikingly, the one participant with PD (NS22) did have the lowest proportion of NSBs (0.15) of the neurogenic stuttering group and the second highest proportion of SLD (0.26). Both participants with TBI (NS16 and NS17) had relatively high severity scores and the participant with ALS (NS21) had the highest mean severity score. These groups are, however, too small reliably give evidence on the influence of aetiology on non-speech behaviours. More research on the characteristics of non-speech behaviours and neurogenic stuttering in different aetiologies is needed.

In a second analysis, the results to the SSC-ER and BCL were added as possible predictors in a regression with sixteen participants (72%) of the neurogenic stuttering group. However, a relationship between the questionnaires and the proportion of NSB or the combined NSB value was not found. This is, again, surprising, as it is often cited that non-speech behaviours develop as a reaction to stutters (Guitar, 1998; Van Riper, 1971). It seems very intuitive that participants with severe emotional reactions to their speech develop more movements or tension while speaking. The cases of neurogenic stuttering that were described in the literature as anxious also did not show many non-speech behaviours. The 55-year old female

described by Nowack and Stone (1987), although annoyed an anxious about her speech, did not show any non-speech behaviours. The 36 year-old male in the study of Attanasio (1987) did have anxiety about his speech, but the only reported non-speech behaviour was 'tension'. However, there are also reports of people with neurogenic stuttering with both severe emotional reactions and a lot of non-speech behaviours, for example the 65 year-old female described by Bijleveld et al. (1994). Contrastingly, of the studies reporting cases with non-speech behaviours that also reported emotional reactions, three cases reported only mild emotional reactions such as 'aware but not desperate' (Lebrun et al., 1990), 'annoyed' (Theys et al., 2009) and 'concerned' (Vanhoutte et al., 2014). The difference in all reports in the literature show that the population of people with neurogenic stuttering is heterogenic, which is supported by the data in this study. Of the four people that scored above the cut-off score on the SSC-ER, two (NS21 and NS16) had a high mean severity of NSBs, and one (NS22) had relatively low NSB proportions and severity measures but a high mean NSB duration. NS3 had low overall NSB and disfluency measures.

The BCL did not directly investigate emotions and attitudes about stuttering, but rather the amount of strategies the participants used to aid them with their speech. These strategies vary from getting out of speaking situations by for example "pretending not to know the answer" but also include non-speech behaviours such as "touching your hair", "shaking your head" , "blinking" and "grimacing" (Brutten & Vanryckeghem, 2003) . Surprisingly, the BCL was not a predictor of proportion of NSBs or the combined value of NSBs. Participants with high NSB measures did not always have high scores on the BCL, which is shown in appendix B. NS4 and NS20 for example, had high NSB measurements but a low score on the BCL. This suggests that participants were not always aware of the NSBs. Another explanation is that these participants were aware of the NSBs, but that these NSBs were not conscious strategies they used to aid them with their speech but rather intrinsic symptoms to their stuttering disorder. In contrast, NS9 and NS7 had high scores on the BCL but low NSB measurements. An explanation is that these participants might predominantly use strategies that were not measured in this study, for example "speaking rhythmically" or "changing a word for another word".

In conclusion, the results in this study indicate that the proportion of NSBs and the combined NSB value combining proportion, duration and severity of NSBs, might be influenced by a number of factors. It seems clear, however, that proportion of SLD and proportion of OD do predict the proportion and combined NSB value. This indicates that the occurrence of NSBs might be complex and influenced by different factors.

7.3 Influence of outliers

The range was wider in the neurogenic stutter group in proportion and duration of the disfluencies, indicating that there is more variation between participants within the neurogenic stuttering group compared to the control group. This is to be expected since a wide range of variation is often found in clinical populations. The proportion of SLDs and ODs also varied strongly within the neurogenic stuttering group. This indicates that studying characteristics of

neurogenic stuttering is most effective within a bigger participant group rather than in case-studies to receive a full picture of non-speech behaviours.

There were some participants, however, who could be considered outliers even in the varied neurogenic stuttering group as their scores differed more than two standard deviation from the mean score. These were included in the analysis as their scores are still representative of the population of people with neurogenic stuttering. However, these outliers had an effect on the outcome of the analyses. Excluding NS10 from the analysis with proportion NSB as a dependent variable resulted in a model where proportion OD was no longer a significant predictor. As can be seen in appendix B, NS10 had a high proportion of OD, which could be attributed to neurogenic stuttering, but also to word-finding difficulties and aphasia. The significant relationship between age and severity of NSB might be a result of the high severity scores of NS21 and NS18 who were relatively young participants. Excluding those participants from the analysis resulted in a model with proportion SLD, but not age, as a significant predictor of NSB severity. As this model had a higher adjusted R squared compared to the model including NS21 and NS18 this model might be a better fit. Future studies on neurogenic stuttering should take into account that, although outliers are natural occurrences within the neurogenic stuttering population, they could influence statistical analysis.

7.4 Limits and qualities of this study

This is the first study on non-speech behaviours in neurogenic stuttering in a larger group of participants. As most of the literature consists of case-studies, of which only half report on non-speech behaviours, this study gives a unique insight on non-speech behaviours in neurogenic stuttering. The non-speech behaviours were systematically annotated, and to provide the most complete and objective measure of non-speech behaviours the amount, duration and severity of NSBs were analysed.

This is also the first study to compare the results of people with neurogenic stuttering, to a healthy control group. The control group consisted of elderly persons, four of which had SLD frequencies over 3%. An SLD frequency of 3% or higher often used as a diagnostic criteria for stuttering. These four participants however, were not diagnosed with (neurogenic) stuttering or any other speech- and language disorder, nor did they report speech and/or language difficulties. Vanopdenbosch (2013), surprisingly, reported that none of the elderly speakers in her thesis had an SLD frequency above 3%. As the control group in this study were the same as the group of elderly speakers Vanopdenbosch (2013) reported on, it is clear that in at least some cases both researchers differed in amount of SLDs that occurred. The 3% SLD frequency as a diagnostic criteria is based on a normative study amongst children (Ambrose & Yairi, 1999; Yairi & Seery, 2015). Future normative studies might investigate the SLD frequency in different age groups and whether a criterion of 3% SLD frequency is sufficient to diagnose (neurogenic) stuttering in older individuals.

Secondly, as the control group consisted of healthy individuals, and the neurogenic stuttering group had neurological damage and co-occurring speech- and language disorders, this

could have had an effect on the results. It would therefore be interesting to have a second control group consisting of participants with similar neurological aetiologies and co-occurring disorders, but no neurogenic stuttering.

The classification of non-speech behaviours was adopted from the SSI-4 (Riley, 2009). Although the list of non-speech behaviours in the SSI-4 is not meant to be a classification for non-speech behaviours, there were several reasons for choosing this as a classification. Firstly, there is a lack of official classification systems for non-speech behaviours. The SSI-4, a diagnostic instrument for stuttering, was chosen because this is an instrument that is well-known and used internationally. However, it is possible that this classification is not the best suited classification for non-speech behaviours in neurogenic stuttering. Some behaviours were not seen in any of the participants, for example whistling. Other behaviours could not be classified using the SSI-4 classification, and had to be annotated as "other movements". Head movements like nodding of the head, could not be classified under any of the head movements and were therefore annotated as "other head movements". Of all non-speech behaviours, 52% were marked as "other head movements". It is unknown whether the SSI-4 classification can be used as non-speech behaviours. Now that it is shown that NSBs are present in people with neurogenic stuttering, future studies should focus on identifying the types of NSBs that are associated with neurogenic stuttering, and whether the NSBs that occur in people with developmental stuttering and neurogenic stuttering differ.

It was for the analysis of the videos not possible to have a blind analysis, therefore the person that analysed the videos knew whether or not that person was diagnosed with neurogenic stuttering. There are several reasons why blind analysis was not possible. Firstly, the amount of SLDs was obvious for a speech-language pathologist in most people with neurogenic stuttering. Some of the people with neurogenic stuttering also had other speech- and language problems which were also noticeable for the researcher. Secondly, half of the videos was recorded in a hospital setting, and most conversations included a case-history of the disease. Lastly, the persons interviewing the neurogenic stuttering group were different from the person interviewing the healthy control group.

Although some studies suggest that reliability of stuttering measurements are generally low, it is possible to achieve high levels of interrater reliability in studies that measure stuttering behaviours (Yaruss, 1997). The correlation between the analyses of two raters were high on the proportion of SLD, NSB and OD. The correlation of duration of NSBs was very low. Strikingly, there was a high correlation on severity measures of NSB, indicating that the two raters gave roughly the same severity scores to the NSBs of the participants. This is surprising, as severity of the NSBs would seem a more subjective measure of NSBs as opposed to the duration of NSBs. A closer look at the analyses of both raters reveals several explanations for the low correlation on duration of NSBs. Although both raters mostly annotated movements at roughly the same moment, there were differences in the amount of movements that were scored. One rater could score movements as one longer movement (for example *other head movements*), whereas the

other rater scored it as two separate, shorter, movements (for example *turning away* and *other head movements*). These disagreements influence both proportion and duration of NSBs. When both raters scored the same movement, they did not always agree on duration of the movement. In conclusion, scoring movements during speech is complex. However, both raters scored the movements similarly on all measurements except for duration of NSB. In future studies that study non-speech behaviours that want to include NSB duration, it is important to address this interraterreliability. Overall, the intraraterreliability was high as well. This indicates that the first rater scored all movements consistently throughout this study.

7.5 Conclusion

This is the first study to investigate non-speech behaviours (such as eye blinking, nodding and grimacing) in a larger group of people with neurogenic stuttering. This study aimed to investigate whether people with neurogenic stuttering present with more, longer and/or severe non-speech behaviours compared to fluent speakers. The results show that participants of the neurogenic stuttering group had significantly higher proportions of NSBs and that these NSBs were also more severe and longer in duration. This shows that people with neurogenic stuttering can present with NSBs, contradicting the often cited characteristics of neurogenic stuttering that people with neurogenic stuttering do not present with NSBs. The second aim was to investigate whether the stuttering frequency predicts the amount, duration and severity of these NSBs. The analysis showed that proportion of SLD did significantly predict proportion and duration of NSB, as well as a combined NSB score. Proportion of OD was also an important predictor of NSBs. This indicates that people with more severe neurogenic stuttering also present with more NSBs compared to people with mild neurogenic stuttering. The last aim was to analyse possible predictors of NSBs within the neurogenic stuttering group. Time post-onset, emotions and attitudes about speech and co-occurring speech- and language disorders did not significantly predict NSBs, contradicting the theory that NSBs develop as a reaction to stuttering. However, longitudinal follow-up of participants is needed to further investigate this factor. Future studies should also investigate whether different aetiologies result in differences in non-speech behaviours. Additionally, further research is needed to study the other characteristics proposed by Helm-Estabrooks (1999) in a larger group of neurogenic stuttering participants. Finally, speech- and language therapists working with people with neurogenic stuttering need to be alert that both non-speech behaviours and emotions and attitudes about speech may be present and should be addressed.

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Appendix A: Overview of all participants included in the study

Table 7, overview of aetiology, age, gender and time post-onset of participants in the neurogenic stuttering group and age and gender of participants in the control group

Neurogenic stuttering group					Control group		
Participant number	Medical aetiology	Age	Gender	Time post-onset (days)	Participant number	Age	Gender
NS1	CVA	71	M	9	C1	71;04	F
NS2	CVA	86	M	53	C2	72;08	M
NS3	CVA	51	M	26	C3*	73;06	F
NS4	CVA	60	F	8	C4	73;09	F
NS5	CVA	76	M	131	C5*	74;02	F
NS6	CVA	48	F	518	C6	74;05	F
NS7	CVA	72	M	75	C7	74;11	M
NS8	CVA	79	F	211	C8	77;00	M
NS9	CVA	84	F	190	C9	77;07	M
NS10	CVA	61	M	91	C10*	79;01	M
NS11	CVA	73	F	2	C11	80;05	M
NS12	CVA	67	F	107	C12	80;11	F
NS13	CVA	76	M	5	C13	81;05	F
NS14	CVA	79	M	12	C14	82;09	M
NS15	CVA	75	M	6	C15	83;06	M
NS16	TBI	46	M	78	C16	85;05	M
NS17	TBI	67	F	27	C17	86;01	F
NS18	CVA	50	M	365	C18	86;07	M
NS19	CVA	56	F	74	C19	92;01	F
NS20	CVA	63	M	1274	C20	92;06	F
NS21	ALS	52	M	180			
NS22	PD	58	M	330			
NS23*	CVA	76	M	NA			
NS24*	CVA	70	M	12			
NS25*	CVA	61	M	NA			
NS26*	CVA, Dementia	88	F	10			
NS27*	CVA	82	M	7			

*Excluded from analysis due to speech sample smaller than 200 words

Appendix B: Overview of all variables per participant

Table 8, overview of all variables (gender, age, time post-onset, number of co-occurring speech- and language disorders, aetiology, SSC-ER score, SSC-ED score, BCL score, S-scale score, proportion of SLDs, proportion of ODs, proportion of NSBs, per participants, mean severity score, number of NSBs with a severity score of resp. 1, 2, 3 and 4, mean duration of NSBs and the combined NSB score) per participant.

Participant number	Gender	Age	Time post-onset (days)	Number of co-occurring disorders	Aetiology	SSC-ER score	SSC-ED score	BCL score	S-scale score	Proportion of SLDs	Proportion of ODs	Proportion of NSBs	Mean severity score	Nr. Of NSBs with a severity score of 1	Nr. Of NSBs with a severity score of 2	Nr. Of NSBs with a severity score of 3	Nr. of NSBs with a severity score of 4	Mean duration of NSBs (ms)	Combined NSB score
C1	F	71								0,033	0,083	0,200	1,000	44	0	0	0	1273	0,013
C2	M	72								0,043	0,040	0,277	1,044	87	4	0	0	1059	0,018
C4	F	73								0,017	0,063	0,250	1,200	60	15	0	0	1386	0,019
C6	F	74								0,010	0,023	0,303	1,055	86	5	0	0	1363	0,021
C7	M	74								0,010	0,030	0,183	1,036	54	0	1	0	1139	0,012
C8	M	77								0,013	0,023	0,180	1,037	52	2	0	0	1306	0,012
C9	M	77								0,000	0,017	0,203	1,148	52	9	0	0	1868	0,015
C11	M	80								0,037	0,077	0,250	1,093	68	7	0	0	889	0,017
C12	F	80								0,013	0,057	0,107	1,000	37	0	0	0	1996	0,007
C13	F	81								0,010	0,023	0,103	1,065	29	2	0	0	1989	0,008
C14	M	82								0,010	0,070	0,120	1,059	32	2	0	0	1257	0,008
C15	M	83								0,017	0,070	0,087	1,000	26	0	0	0	1283	0,005
C16	M	85								0,010	0,013	0,150	1,043	44	2	0	0	1094	0,010
C17	F	86								0,003	0,047	0,150	1,250	27	9	0	0	1295	0,012
C18	M	86								0,010	0,030	0,117	1,059	32	2	0	0	1268	0,008
C19	F	92								0,040	0,037	0,177	1,019	52	1	0	0	1162	0,012
C20	F	92								0,020	0,020	0,187	1,161	49	5	2	0	1509	0,014
NS1	M	71	9	2	CVA	-	-	-	-	0,063	0,023	0,290	1,161	74	12	1	0	1909	0,022
NS2	M	86	53	0	CVA	51	104	1	15	0,203	0,063	0,260	1,214	55	15	0	0	1488	0,020
NS3	M	51	26	1	CVA	137	110	-	25	0,041	0,046	0,228	1,040	48	2	0	0	1237	0,015
NS4	F	60	8	2	CVA	68	-	9	22	0,393	0,092	0,519	1,429	67	31	7	0	2627	0,052
NS5	M	76	131	1	CVA	54	52	2	6	0,030	0,027	0,157	1,196	44	5	1	1	1108	0,011
NS6	F	48	518	0	CVA	68	74	12	13	0,020	0,020	0,073	1,045	21	1	0	0	1044	0,005
NS7	M	72	75	0	CVA	114	180	31	35	0,037	0,047	0,093	1,000	17	0	0	0	1669	0,006
NS8	F	79	211	0	CVA	122	109	14	29	0,100	0,040	0,263	1,090	61	6	0	0	2021	0,019
NS9	F	84	190	1	CVA	105	58	29	18	0,050	0,010	0,160	1,149	40	7	0	0	2398	0,013
NS10	M	61	91	2	CVA	103	123	151	18	0,545	0,709	0,991	1,387	105	61	2	0	2186	0,091
NS11	F	73	2	0	CVA	57	51	10	15	0,147	0,157	0,333	1,247	65	12	4	0	1598	0,026
NS12	F	67	107	0	CVA	59	58	0	19	0,063	0,063	0,173	1,045	42	2	0	0	2012	0,012
NS13	M	76	5	1	CVA	56	55	-	8	0,043	0,073	0,300	1,144	77	13	0	0	1255	0,022
NS14	M	79	12	1	CVA	56	57	46	21	0,170	0,153	0,297	1,174	58	10	1	0	1688	0,023
NS15	M	75	6	2	CVA	-	-	-	-	0,044	0,019	0,189	1,051	37	2	0	0	1267	0,012
NS16	M	46	78	2	TBI	129	117	18	21	0,080	0,060	0,223	1,333	43	9	5	0	2054	0,020
NS17	F	67	27	1	TBI	53	51	5	12	0,140	0,090	0,313	1,257	54	14	2	0	1972	0,026

NS18	M	50	365	0	CVA	-	111	41	20	0,103	0,143	0,290	1,987	29	21	24	2	1116	0,036
NS19	F	56	74	0	CVA	-	-	-	-	0,030	0,023	0,310	1,189	73	17	0	0	1424	0,023
NS20	M	63	1274	1	CVA	104	98	31	22	0,050	0,070	0,293	1,060	63	4	0	0	1672	0,020
NS21	M	52	180	1	ALS	133	182	21	16	0,100	0,053	0,210	1,812	32	38	14	1	5653	0,041
NS22	M	58	330	1	PD	181	175	19	29	0,260	0,063	0,157	1,111	40	5	0	0	2277	0,012