Stuttering and Speech-Rhythm

Inaugural-Dissertation
zur Erlangung des Doktorgrades
der Philosophie an der Ludwig-Maximilians-Universität
München

vorgelegt von
Ramona Schreier

2024

Erstgutachter: Prof. Dr. Phil Hoole

Zweitgutachterin: Prof. Dr. Simone Falk

Tag der mündlichen Prüfung: 21. Februar 2022

Content

List of Abbreviations

AF	arcuate fasciculus
BG	basal ganglia
CB	cerebellum
CV	coefficient of variation
CWS	children who stutter
CWNS	children who do not stutter
DIVA	Directions into Velocities of Articulators
DTI	Diffusion Tensor Imaging
FLS	fasciculus longitudinalis superior
GODIVA	Gradient Order Directions into Velocities of Articulators
IOI	inter-onset-interval
IVI	inter-vowel-interval
MRI	Magnetic Resonance Imaging
NMA	negative mean asynchrony
OASES	Overall Assessment of the Speaker’s Experience of Stuttering
PMC	premotor cortex
PWS	persons who stutter
PWNS	persons who do not stutter
SMA	supplementary motor area
SMS	sensorimotor synchronization
SSI-3	Stuttering Severity Instrument – Third edition
STG	superior temporal gyrus
VOT	voice onset time

Abbreviations in figures 2.4, 2.5 and 2.6 are specified in the respective legends below the figure

List of Figures

List of Tables

1. Introduction

„One of the hardest things in life is having words on your heart that you can't utter. "

This quotation by the famous American actor James Earl Jones (Pine, 2011, p. 107), who was affected by stuttering himself, sums up one of the major problems persons who stutter (PWS) are confronted with every day. Although PWS exactly know what they want to say, they find themselves unable to produce the words in the moment of articulation (Guitar, 2014). Throughout all different cultures and languages, about one per cent of the adult population is affected by stuttering, (Yairi & Ambrose, 2013). Despite intense research on this topic over the last decades, the actual causes of stuttering remain unclear. Due to its high variability and heterogeneity many theories and models have tried to account for or offer a cure to this phenomenon – but none of them can fully explain the origin nor heal the symptoms of stuttering in all persons affected (Van Riper, 1982; Ward, 2006). Over the last years, brain structure and function have been examined more closely as possible sources. In the research on the causes and neurological underpinnings of stuttering, a great deal of progress has been made in finding differences between people who do and people who do not stutter. However, there is still no answer about the role those aberrations play in stuttering (Etchell, Civier, Ballard, & Sowman, 2018): Are they of causal or rather of compensatory nature?

One fact that is currently a matter of great interest is the link between stuttering and timing abilities. Not only for PWS but also in persons affected by developmental dyslexia or specific language impairment there seems to be a link to rhythmical abilities, which were shown to be impaired in this population (Flaugnacco et al., 2014; Jentschke, Koelsch, Sallat, & Friederici, 2008). The abilities of processing rhythmical information or producing rhythm – in speech or in any other motor action - are very well understood in fluently speaking persons (Ravignani, Honing, & Kotz, 2017). When it comes to populations with specific impairments of language or speech, like stuttering, there are still many unanswered questions about the exact type of disruption in these processes during speech perception or production. Various studies have demonstrated that rhythmical abilities and the timing of motor actions – including speech – seem to work in a different way or seem to underlie different timing mechanisms in stuttering speakers compared to fluently speaking persons (Etchell, Johnson, & Sowman, 2015; Falk, Müller, & Dalla Bella, 2015; Max & Yudmann, 2003; Olander, Smith, & Zelaznik, 2010). These recently found results combined with the established methods leading to a more fluent speech in stuttering speakers, like choral or paced speech, suggest that exactly those methods seem to make use of an external rhythm, which have shown to lead to more fluent speech in PWS (Davidow, 2014; Etchell, Johnson, & Sowman, 2014). Furthermore, this enhanced fluency under fluency inducing conditions was also shown on neuronal levels with the help of imaging techniques (Toyomura, Fujii, & Kuriki, 2011). Those well-known effects of fluency-inducing conditions along with the latest results of studies on brain imaging techniques in stuttering speakers all point to the assumption that PWS seem to be affected by a rhythmical deficit. However, we do not know so far where exactly deficient mechanisms are located or even whether it is a problem of perception or rather one of production. Studies on this topic were able to prove that PWS show a greater variability in tasks of rhythm production – mostly tested with finger tapping or hand clapping tasks (Falk et al., 2015; Max & Yudmann, 2003; Olander et al., 2010). More than three decades ago, Harrington (1988) proposed a model of speech production in stuttering speakers. The key proposition in this thesis is the possible explanation of disfluencies as a result of a timing deficit, which can be characterized as an erroneously-perceived asynchrony in one's own speech production leading to an interruption of the onward flow of speech due to maladaptive processes in the timing of articulatory movements. So far, no study was actually able to prove this proposition to be correct for the speech production of PWS, but some studies were able to show that there seems to be an aberrant way of processing rhythmic structures in the stuttering speakers' perception (Wieland, McAuley, Dilley, & Chang, 2015). Considering what is already known about the different mechanisms that seem to underlie the perception and production of rhythm in stuttering versus in fluently-speaking persons the question arises whether stuttering speakers do have a general deficit in the rhythmical patterning of speech and whether the audible and visible symptoms of stuttering are a result of this exact problem.

To come closer to answering this question the present thesis aims to investigate the link between stuttering and rhythm in perception and production in a population that has been investigated rather sparsely so far: children and adolescents. The adult stuttering population has undergone many behavioral and brain-imaging studies regarding this question – but at the same time an adult stuttering person has adapted to his or her stuttering over the years and maybe also shows certain strategies of compensation whereas children and adolescents, who do not have such a long experience in coping with or adapting to their symptoms, are the more interesting population to look at regarding the origin of stuttering symptoms. Comparing those results to a normally-speaking group of age-matched control participants, new insights into the verbal perception and production of rhythm can be gained. The aim of these investigations is not only a better understanding of the origin of stuttering symptoms but also - in the long run – an improvement of therapeutic intervention.

The thesis at hand is structured as follows: After the introduction, a comprehensive theoretical background is given in the second chapter. Chapters 3, 4 and 5 contain the three studies conducted: Whereas in Chapter 3 speech rhythm is being investigated in a very basic form, namely speech and articulation rate, chapter 4 is concerned with more detailed rhythmical abilities, as participants are asked to verbally synchronize to a given rhythm. Finally, within chapter 5, the link between rhythmical abilities and therapeutic outcome is investigated: Can rhythmic abilities, as assessed within chapter 3 and 4, predict therapeutic outcome? This question is extremely relevant for further therapeutic interventions. In chapter 6, all results are summed up and discussed in the light of current literature on this topic.

In chapters 3, 4 and 5, within the section on participants, repetition on participant acquisition and compilation was caused by the fact that groups differed slightly due to various comorbidities that made participation impossible for one study but did not interfere with the question of another study within this thesis.

2. Stuttering

This chapter is supposed to give a comprehensive overview of the disorder and its relevant aspects ensuring a proper understanding of the questions raised and the hypothesis postulated. Furthermore, a precise definition of stuttering as well as its possible causes and symptoms will be given.

2.1 Definition of stuttering and speech fluency

Defining stuttering is – although it is one of the most commonly known disorders of speech – quite difficult (Natke & Alpermann, 2010). For one thing, it is not a homogenous clinical picture but rather a syndrome consisting of a combination of symptoms of motoric, psychosocial, and linguistic symptoms. Additionally, it is - despite years of research – not easy to determine whether a child shows symptoms of stuttering or normal disfluency (Howell, 2011; Ochsenkühn, Frauer, & Thiel, 2015). When using the term normal disfluency, one must define fluency in speaking, which is hard to define itself (Guitar, 2014).

In order to define disfluency, one has to specify what fluency is first and then identify deviations from it: Levels of fluency can vary extremely from one speaker to another, and no person can ever speak completely fluently. Every speaker – even the most eloquent ones – will more or less frequently produce speech errors, which can be a range of hesitations or phrase revisions; but also repetitions of words or the use of interjections are common (Ward, 2006). Still, listeners would not classify those kinds of disfluencies as pathological or perceive the speaker as a person affected by stuttering. Starkweather proposed that it is mostly temporal aspects that determine fluency in speech production (Starkweather, 1980, 1987). Pauses, intonation, stress as well as speech rhythm in general emerge through our temporal control of speech structures. He therefore noted that the flow of information, and not the flow of speech sounds, is one very important factor of fluency. Furthermore, in his opinion, the effort of a speaker is also a major aspect of describing fluency: Not only the physical work that is done during the act of speaking but also the mental work must be considered when speaking about fluency or disfluency (Starkweather, 1987). To sum up, fluency can be described as an "effortless flow of speech" (Guitar, 2014, p. 7). However, even persons speaking fluently show disfluencies in their speech, which must be distinguished from disfluencies that are characteristic of stuttering.

The childhood-onset fluency disorder, as stuttering is called in the fifth Diagnostic and Statistical Manual of the American Psychiatric Association (2013), is the most common type of stuttering and is also referred to in literature as *developmental stuttering* (Ward, 2006). It is listed under communication disorders and its name as seen above was only changed from the fourth to the fifth edition (Cohen, 2014). Also, in the International Classification of Diseases (ICD-10) it is mentioned under behavioral and emotional disorders with a usual onset in childhood or adolescence (WHO, 2016). Due to the difficulties in the definition and diagnosis of stuttering, the DSM-V tries to give a list of precise diagnostic criteria that must be met in order to diagnose the childhood-onset fluency disorder. The first diagnostic criterium is the presence of disturbances of the normal flow of speech inappropriate for age and skills of the child and the persistence of those over time. A list of symptoms that can frequently occur contains repetitions of sounds and syllables, prolongations of consonants and vowels, pauses within words, audible or silent blocks, substitution of words, also called circumlocutions, physical tension during word production and word repetitions of monosyllabic words. The second criterium is the presence of anxiety caused by the symptoms listed above, which can lead to limitations in communication, participation, or performance in professional or academic contexts. Either only single symptoms of the ones listed can occur or a combination of them. The third criterium is the onset in the early developmental age and the fourth excludes any symptoms caused by neurologic insults or other mental disorders (American Psychiatric Association, 2013) . This definition of the fluency disorder is in line with the well-respected definition Wingate gave in 1964, which has served as a kind of standard definition since:

> “The term ‘stuttering’ means:
> 1. (a) Disruption in the fluency of verbal expression, which is (b) characterized by involuntary, audible or silent, repetitions or prolongation in the utterances of short speech elements, namely: sounds, syllables and words of one syllable. These disruptions (c) usually occur frequently or are marked in character and (d) are not readily controllable.
> 2. Sometimes the disruptions are (e) accompanied by accessory activities involving the speech apparatus, related or unrelated body structures, or stereotyped speech utterances. These activities give the appearance of being speech-related struggle.
> 3. Also, there are not infrequently (f) indications or report of the presence of an emotional state, ranging from a general condition of ‘excitement’ or ‘tension’ to more specific emotions of a negative nature, such as fear, embarrassment, irritation, or the like. (g) The immediate source of stuttering is some incoordination expressed in the peripheral speech mechanisms; the ultimate cause is presently unknown and may be complex or compound.” (Wingate, 1964, p. 488)

Comparing this standard definition to the most recent definition of the American Psychiatric Association, one can see that the most important aspects of this fluency disorder were defined precisely already over 50 years ago. Wingate did not yet mention the early onset or the absence of other neurological or mental causes, and he also left out the limitations in participation. Besides that, his definition managed quite well to include all the aspects relevant for the diagnosis of this complex fluency disorder. His definition as well as the one in DSM-V tries to capture all aspects of stuttering, whereas other definitions were rather psychologically based or symptom-led. Going back in time, there was a vast number of definitions, all trying to explain stuttering based on the knowledge available at that time. The oldest definition goes back to Aristoteles (384-322 b.c.), who defined stuttering as an inhibition of the voice due to a malfunctioning tongue (Wirth, 2000). Since then, a lot of research and experience led to the definition that is in use today capturing both distinct elements: the audible and observable features of stuttering symptoms as well as its psychological and social consequences of the individual dealing with the disruptions (Ward, 2006).

2.2 Symptomatology

As mentioned in the previous chapter, stuttering is a complex and individually distinct disorder. It has an extremely variable and diverse appearance, which is why no stuttering person can be compared to another. Even within one individual, stuttering can vary considerably due to temporal and situational conditions (Guitar, 2014; Howell, 2011; Ward, 2006). However, there are certain symptoms that appear in every affected person’s speech, always characterized by a disruption in the temporal flow of speech production units such as sounds, syllables, segments and words (Bosshardt, 2010).

In the German as well as in the English-speaking specialist literature, a classification of stuttering symptoms in “primary and secondary stuttering” (Ward, 2006, p. 6) or in “core” and “secondary behavior” (Guitar, 2014, p. 4) is used to describe symptoms more precisely and systematically. While both terms are used synonymously in the relevant literature, this thesis uses the terms of *core* and *secondary behavior* to describe symptoms in stuttering speakers. Furthermore, symptoms can also be classified as *overt features* that are instantly recognized by the environment, since they are clearly audible and visible, and *covert reactions*, that stand for all emotions, feelings and attitudes that are not directly observable but still happening inside the person affected (Natke & Alpermann, 2010). This classification harks back to van Riper (Van Riper, 1982). Ultimately, this very precise separation and classification is not always as salient as theory might imply, but rather characterized by smooth transitions (Weikert, 2014).

2.2.1 Core behaviors

The term *core behaviors* goes back to Van Riper (1971, 1982), who made it a common term in stuttering literature by describing the basic audible features of the disruption of speech in stuttering speakers. These involuntary interruptions are relevant for the diagnosis of stuttering and can be divided into three categories: repetitions, prolongations and blocks (Guitar, 2014; Van Riper, 1971, 1982). Repetitions can either affect simply a sound (e.g., “I-I-I-I want to…”), a syllable (e.g., “I wa-wa-wa-want to...”), or a single-syllable word (e.g., “I want-want-want to…”), which is then repeated various times. This gives the impression that the speaker is “stuck” (Guitar, 2014, p. 8) on that specific sound, syllable or word and keeps repeating it until the upcoming sound can be articulated. Concerning the repetition of whole words, some authors were not sure about its role, especially in children: Since typically developing children also frequently show repetitions of whole words, there was some disagreement about the symptom-specific character of those repetitions in stuttering (Ambrose & Yairi, 1999; Howell, 2011). Some authors regarded those repetitions as problems in speech planning, lexical retrieval or an immature syntactic system instead (Clark & Clark, 1977; Rispoli, 2003). Nowadays, the repetition of whole, single-syllable words is regarded as a core symptom of stuttering, especially if there is a sign of tension during the repetition or the repetition is articulated at a high rate (Ambrose & Yairi, 1999; Ochsenkühn et al., 2015). Moreover, repetitions are the core behaviors observed the most among those children who are just starting to stutter, and those children who are actually stuttering will repeat the concerned word or syllable quite likely more than twice (Yairi, 1983).

Prolongations are distinctly audible interruptions of the onward flow of speech. They are characterized by the static persistence of the articulators with a simultaneously continuing sound or air flow (e.g., “I wwwwwant to…”). Prolongations can vary significantly in their duration, with prolongations shorter than half a second not even always being perceived as such, but in some cases they may even last longer than one minute (Guitar, 2014; Van Riper, 1982). Affected persons struggle to manage the transition from one sound to the other, which is why they are prolonging the first sound hoping to complete the transition to the following sound (von Tiling, 2012). In contrast to the third core behavior - the block - the prolongation is characterized by the freezing of the articulatory movements but not by the interruption of the sound or air flow (Van Riper, 1982; Wingate, 1964). Therefore, the term prolongation, as it is being used in today’s literature, denotes an ongoing sound or air flow with stopped articulators (Guitar, 2014; Natke & Alpermann, 2010; Ochsenkühn et al., 2015).

Blocks – the third symptom of the core behaviors – are typically the last of the three core behaviors to appear (Guitar, 2014). They emerge all of a sudden as an inappropriate stop of the air and voice flow, mostly accompanied by a stop in the articulatory movements. The mostly silent interruptions of the speech production (e.g., "I want ---- to…") often coincide with a high tension in the articulatory muscles or even other parts of the body. Since they may involve different levels of speech production, they can be divided into different kind of blocks: Often, the air flow is disrupted already at the glottal level, leading to an immediate stop of voicing, which is why this kind of block is sometimes called a *laryngeal block*. However, the interruption of the flow of speech can also be caused by any other constriction in the articulatory system. In this case, one would speak of an *articulatory block* (Natke & Alpermann, 2010). In most cases, any kind of articulatory gesture is being interrupted during a block. When articulating plosives for example, a block can lead to a repetition of the sound when the tension is being eased repetitively. Hence, the audible symptom resembles a repetition, although the underlying mechanism stems from a completly different cause (Zückner, 2008). Blocks often get more tense and longer as stuttering persists. Sometimes tremors may accompany a severe block. Tremors are rapid oscillations which can be observed in the jaw or lips during a symptom. (Van Riper, 1982). Blocks are often experienced as the worst loss of control, since they might sometimes last as long as a few minutes (Guitar, 2014; Natke & Alpermann, 2010; Sandrieser & Schneider, 2008). Since blocks and prolongations are both characterized by a fixation of the articulatory movements, they might be summarized in the same symptom category. Depending on the category of the sound a speaker is trying to articulate, the result can either be silent, e.g., a block, or audible, e.g., a prolongation (Natke, Sandrieser, Pietrowsky, & Kalveram, 2006).

People who stutter vary greatly in the frequency and duration of their symptoms. Stuttering is diagnosed if at least three percent of the spontaneous speech contains these symptoms (Ambrose & Yairi, 1999). Research indicates that the average frequency is about ten percent of the words when reading aloud with a great individual variation (Bloodstein, 1944; Bloodstein & Bernstein Ratner, 2008), but there are also many people affected by stuttering who stutter on five percent of the words or even fewer when speaking or reading aloud. On the other side of the spectrum, some people who stutter do so on more than 50 percent of the words. The duration of the core behavior can also vary significantly but not as much as the percentage of stuttering symptoms itself: They rarely last longer than five seconds and average around one second (Bloodstein, 1944; Bloodstein & Bernstein Ratner, 2008).

2.2.2 Secondary behaviors

Core behavior is almost always accompanied by the so-called secondary behavior that must be distinguished clearly from the first. This part of stuttering emerges in the development of stuttering as an individually learned reaction to the core behavior. The dynamic process of developing secondary behavior is very appropriately summarized by Guitar (2014):

> "People who stutter don't enjoy stuttering. They react to their repetitions, prolongations, and blocks by trying to end them quickly if they can't avoid them altogether. Such reactions may begin as a random struggle but soon turn into well-learned patterns." (Guitar, 2014, p. 9)

The desperate need of speakers affected to regain control over their speech fluency leads to individually very distinct reactions and forms of secondary behavior which led Van Riper to say that there are "literally thousands of possible reactions that can be used to escape, avoid or disguise the inability to say a word" (Van Riper, 1971, p. 126). As time passes, those reactions accompanying the disfluencies are barely controllable and can be divided in two classes: escape and avoidance behavior (Guitar, 2014; Natke & Alpermann, 2010).

2.2.2.1 Escape behavior

The most common reaction to core symptoms is the tension of muscles involved in the process of speaking. A sudden muscle contraction can end a repetition or a prolongation effectively which makes the contraction very functional in the first place and therefore provides a rewarding experience and an emotional relief. The usage of a muscle contraction therefore is positively rewarded by ending the symptom. Usually, the amount of tension grows over time, resulting in a strong contraction of muscles or even a tremor, which is then itself a loss of motor control. Those contractions frequently occur in antagonistic muscles within the mandible. Due to the increased muscle tension while speaking, Bloodstein named this phenomenon *struggle behavior* (Bloodstein, 1958). This struggle behavior relates to any struggle with the intention of initiating or ending stuttering symptoms.

Elevated muscle tension does not only occur in speaking-related muscles but also in other parts of the body. Typically, those head or body movements happen spontaneously, and PWS often are not aware of them happening. A lot of those movements were functional once since they ended a symptom. Therefore, they start as an instrumentally conditioned reaction but lose their functionality in the course of time (Guitar, 2014; Natke & Alpermann, 2010). Those movements can occur in the fine motor skills as well as in gross motor skills: Common examples in the field of escape behavior are eye blinks, frowning, head nods, grimacing, stamping with the foot, clenching of the fist, or interjections of sounds ("uh", "ah" etc.). Since they often happen to terminate a stutter, they are reinforced in the course of time. In case of long-lasting repetitions, prolongations or blocks, PWS react differently to end those symptoms: An increase of sound level or a change in phonation type are typical examples (Van Riper, 1982).

2.2.2.2 Avoidance behavior

In contrast to escape behavior, avoidance behavior is a reaction to the anticipation of core symptoms, meaning that they are learned to prevent the occurrence of stuttering symptoms. To avoid the symptom itself and, of course, the negative emotions it entails, behaviors that previously helped to end symptoms are reused or new behaviors, such as changes in the actual planning of the sentence and the reconstruction of words used, are invented. Avoidance behavior can therefore be either verbal or non-verbal.

Verbal avoidance behavior includes the rephrasing or rearranging of words, phrases or sentences if they contain words that are associated with stuttering within a speaker. Also, they can be replaced by synonyms or descriptions to avoid those specific words. Even changing to an accent or changing the speech rate is part of verbal avoidance. This is because all those strategies mentioned above once led to a reduction of symptoms, but then soon turned into well learned patterns that often become more prominent than a stuttering symptom itself. Furthermore, the usage of interjections (e.g., “uh”, “like”), postponements or starters is a very common avoidance behavior, that can lead – in extreme cases – to a meaningless style of speaking, since utterances barely transport the actual communicative intention.

Non-verbal avoidance behavior used to postpone or to skip disfluencies, such as facial movement or body movement, is sometimes even used as timing devices: They are supposed to help making the precisely timed usage of a certain word more predictable (Natke & Alpermann, 2010).

Both verbal and non-verbal avoidance behavior provides a highly rewarding relief from the constant and growing fear that symptoms might occur. However, soon after some supposedly “successful” experiences these avoidance behaviors turn into strong habits that become hard to change and often are used unconsciously (Guitar, 2014).

2.2.2.3 Feelings and attitudes

Covert reactions encompass all the feelings, reactions and attitudes of PWS which are most commonly not observable to outsiders. The feeling most frequently described by stuttering speakers is fear – fear of a social rejection due to negative reactions of the listeners or fear of the loss of control and the inability to communicate. The anticipation of an imminent loss of control is one of the most burdensome factors in stuttering, as this quotation from Van Riper illustrates:

> „The inability to move a muscle when you want to move it, a muscle that you can normally move with ease, is traumatic to the basic integrity of the self. Equally devastating is the experience of being unable to stop doing something that you don't want to do." (Van Riper, 1971, p. 158)

On top of that, PWS not only develop fear, but they are also ashamed about the loss of control. They feel like they put their listener into an uncomfortable position. Those permanently stressful and incontrollable situations also lead to a high degree of frustration, which increases due to ongoing negative experiences about their own speech fluency. The stuttering speaker is in a state of constant anticipation of his/her own failure and this is – from his/her perspective – affirmed over and over again (Ochsenkühn et al., 2015). More than half of the two-year-old stuttering children are already aware of their disfluencies and express this verbally or non-verbally. With higher age the awareness rises up to 90% in seven-year-olds (Boey et al., 2009). Those mental burdens are often accompanied by an enormous psychological strain and can lead to a negative and inferior self-concept. During conversations, those negative attitudes are projected on listeners as the person who stutters automatically assumes he/she is perceived as stupid, insecure and nervous (Boey et al., 2009; Guitar, 2014). These negative feelings can consequently lead to a social withdrawal and have a negative impact on social interaction and social integration (Ochsenkühn et al., 2015).

2.2.3 Variability in stuttering symptoms

Stuttering severity can vary greatly within one speaker depending on the communicative situation. This heterogeneity exists even if only looking at developmental stuttering, as is done in this thesis. Consequently, the diagnosis of stuttering is something that must be done with great caution and various factors must be considered. Those factors will be discussed in 2.4.1. However, in the context of stuttering symptoms it is important to know that there is a great deal of variability within one speaker: there are, on the one hand, contexts and situations that lead to a more or even completely fluent speech and, on the other hand, contexts and situations that might generate more disfluencies. Understanding this feature of stuttering is crucial when it comes to the questions raised in this thesis. Therefore a theoretical background is given. Conditions that can lead to more severe and more frequent symptoms are usually interactive and communicatively demanding situations such as speaking with strangers, speaking on the telephone, conversations with a high communicative responsibility or emotional involvement.

Furthermore, linguistic and speech motor complexity also play an important role here (Bloodstein & Bernstein Ratner, 2008; Peters, Hulstijn, & van Lieshout, 2000).

Situations that typically reduce the frequency and severity of stuttering symptoms are communicatively less demanding situations (e.g., talking to a baby or a pet), singing, choral speech, masked speech, speaking with delayed auditory feedback, speaking in a changed manner of speech (e.g., acting, imitation of dialects/persons), speaking to an external rhythm or automatized speech (e.g., cursing, counting etc.). In sum, it becomes obvious that factors somehow changing the way of "normal" speaking can temporarily lead to an increased fluency. Those situational effects must therefore not be mistakenly interpreted as therapeutic success but as a simple and commonly known effect enhancing speech fluency in stuttering speakers (Bloodstein & Bernstein Ratner, 2008; Hulstijn, Summers, van Lieshout, & Peters, 1992; Natke, Sandrieser, van Ark, Pietrowsky, & Kalveram, 2004; Ochsenkühn et al., 2015).

2.3 Epidemiology

During the last and current century, epidemiological advances in stuttering research have been made. New information on factors determining and influencing the disorder and its presence or absence, its distribution, the frequency it occurs within the general or a certain population, subtypes, remission, and different manifestations it can take has expanded scientific knowledge. In the following paragraphs, these recent advances are systematically depicted and added to existing facts about stuttering. However, since the causes of stuttering are a very complex field, they are will be explored separately in a later chapter (see Chapter 2.5).

2.3.1 Onset

Many studies posing the questions as to when stuttering is most likely to appear and who is at risk of developing it were conducted during the last years. Most of them established clearly that the majority of cases already emerge in childhood, which is why they also most largely agreed on stuttering being a disorder with onset in early childhood (Brocklehurst, 2013; Guitar, 2014; Van Riper, 1982; von Tiling, 2012; Yairi, 1983; Yairi & Ambrose, 1999, 2013). In 50% of all cases, stuttering onset was before the fourth year of life, in 75% it was before the sixth year of life and in 99% of all cases stuttering onset was before the 12th year of life (Andrews, 1985). Although this data seems very unambiguous and was the standard for many years, newer studies showed deviations from these numbers concerning onset data. Those more recent studies conducted in the United Kingdom, Denmark, Australia and the United States. report an even lower age at onset with an average of approximately 33 months (Buck, Lee, & Cook, 2002; Månsson, 2000; Reilly et al., 2009; Yairi & Ambrose, 2005, 2013). According to Yairi and Ambrose (2005), it is the age-span between 24 and 35 months where 60% of all onsets occur. Other authors, however, conclude that there are two distinct groups differing in terms of onset conditions: whereas some children already show a very early onset in the second or third year of life, others have a rather late onset in the fourth or fifth year of life. Authors establish links between those two groups in terms of mechanisms of speech development happening rather simultaneously, as within the first group children who are starting to use longer and more complex linguistic units, whereas within the second group a fluent usage of grammatically complex and correct sentences was already learned (Johannsen, 2009).

The onset of stuttering can happen suddenly or is perceived as gradual by parents and caregivers. While those parents reporting a gradual development of stuttering symptoms mostly cannot give a precise time of onset, there also are parents, often those reporting a very sudden onset of stuttering, that relate this first appearance of symptoms to certain traumatic events, such as the death of a family member or the birth of a sibling (Natke & Alpermann, 2010; von Tiling, Crawcour, & Hoyer, 2014). In general, however, stuttering onset is not related to a specific event (Van Riper, 1982).

2.3.2 Incidence and prevalence

When it comes to analyzing data on incidence and prevalence in stuttering, this is not straightforward. This is because data on incidence, indicating the number of persons newly affected by a disease or a disorder in a certain population within a specified period of time, as well as data on prevalence, indicating the number of persons generally affected at a certain point of time by a disease or a disorder, vary significantly depending on the timeframe of investigation within a certain population (Yairi & Ambrose, 2013). Looking at two- to four-year-olds, both values would be rather close, since at that age the number of newly affected children and the number of spontaneously remitted children are quite balanced. According to latest results in research on incidence, this exact timeframe captures not only the highest number of newly affected children but also the highest number of children recovering spontaneously (Yairi & Ambrose, 2005). For a long time, it was postulated that up to 8% of all children experience a disfluent phase during speech development. Throughout this phase, about 5% of those children show symptoms of early stuttering, hinting towards a chronic development. Based on the fact that four out of five of those children affected would recover during their development, 1% of persons are still affected by stuttering in the adult population (Johannsen, 2009). In terms of incidence, however, it must be mentioned, that the value of 5% over the lifespan that has been assumed for a long time is too conservative. Newer studies presume that the value is around 8% (Brocklehurst, 2013; Månsson, 2000; Yairi & Ambrose, 2013): For the ages of four to five years, incidence varies between 5.04% (Månsson, 2000) and 11.2% (Reilly et al., 2009). In around 40% of affected children, stuttering manifests suddenly, within one to three days, in one third the onset takes about one to two weeks and in the last third, stuttering manifests over a timespan of one to three weeks (Reilly et al., 2009; Yairi & Ambrose, 2005). Prevalence, as already mentioned above, varies depending on the age of the population investigated. The value of prevalence is highest in a population between two and six years, afterwards it decreases. There are two reasons for this fact: Firstly, the phenomenon of spontaneous recovery most commonly takes place within three to four years after onset and therefore tends to happen prior to the age of seven (Yairi & Ambrose, 1999). Secondly, after this age span there are only few onsets of stuttering (Yairi & Ambrose, 2013). In a synopsis of data available a mean prevalence of 1% (span of 0.3 to 2.12) is assumed for children and adolescents aged two to eighteen years (Bloodstein & Bernstein Ratner, 2008). To this day, prevalence data has mostly been collected with school-aged children, therefore data in younger populations is missing. In 18 studies of US American school children a mean prevalence of 1.02% was found, whereas 28 European studies show results of 1.38% of prevalence (Bloodstein & Bernstein Ratner, 2008). A very recent representative collection in Australia showed results comparable to the European studies: a mean prevalence in school aged children of 1.3% - 1.4% was found (Craig & Tran, 2005). Data on prevalence vary due to methodological differences between data collections in terms of the population under investigation (age, gender, and genetic predisposition). In the adult population prevalence is distinctly lower, ranging from 0.8% in males and 0.2% in females (Craig & Tran, 2005).

In summary, it can be stated that incidence and prevalence of stuttering is very dependent on the population under investigation, leading to strongly varying numbers. It is therefore common to calculate mean values of different studies to get closer to the actual number (Natke & Alpermann, 2010).

2.3.3 Gender bias and genetics

What is indisputable, however, is the uneven distribution of gender: significantly more boys than girls are affected by stuttering. In early childhood a ratio of 2:1 or even 3:1 to the disadvantage of boys is reported in literature (Howell, 2011; Johannsen, 2009; Månsson, 2000; Yairi & Ambrose, 1992, 2013). In adults, the gap further grows to a ratio of 4:1 or even 5:1

(Bloodstein & Bernstein Ratner, 2008) due to more frequent recoveries in girls (Craig & Tran, 2005). This phenomenon results in 80% of the adult stuttering population being male. The reason for the change in the gender ratio can be explained by recovery: a significantly higher proportion of girls (spontaneously) recovers from stuttering, leading to a strong bias in the gender distribution. The reason for this imbalance between sexes is not completely known so far; however, genetic factors are assumed to be one of the main causes of this phenomenon. Genetic causes are a popular explanation in this respect since the gender gap exists in all cultures. Furthermore, boys usually show a slower speech and language development with a higher propensity for disorders such as speech sound disorders or dyslexia (Guitar, 2014; Nippold, 2018). Those facts hint towards a gender-specific predisposition (Natke & Alpermann, 2010). Based on these observations, research on genetic causes of stuttering was conducted in family and twin studies. It was clear, that genes play an important role in stuttering: while the concordance, i.e. the corresponding occurrence of certain characteristics, for stuttering was 77% in monozygotic twins, it was only 32% in dizygotic twins and only 20% in normal siblings (Frigerio-Domingues & Drayna, 2017; Howie, 1981b; Kraft & Yairi, 2012). In addition to family and twin studies, there is further genetic research which has gained interest during the last years and supports the idea that genes play an important role in developmental stuttering. Although it is still too early to identify single specific genes relevant for the occurrence of stuttering, research is getting closer to identifying certain genes associated to the condition (Frigerio-Domingues & Drayna, 2017; Kraft & Yairi, 2012; Yairi & Ambrose, 2013).

2.3.4 Race, ethnicity, cultural, and social factors

Stuttering is present in all social classes, all cultures that have undergone investigation about stuttering so far, all nations and all ethnic groups. A synopsis of all epidemiological research published shows, however, that data on incidence and prevalence in different countries, cultures and populations can vary significantly (Bloodstein & Bernstein Ratner, 2008; Proctor, Yairi, Duff, & Zhang, 2008; Yairi & Ambrose, 2013). So far, neither genetic, nor methodological nor cultural explanations can be excluded as reasons for those differences. The shortage of high-quality and credible data in studies of different socio-economic states, cultures, races and ethnicities during the 20th and 21st century is one of the main reasons for the lack of clear facts. Most of those studies suffered from limitations such as failed separation of culture from race or socio-economic status. Furthermore, the effect of bilingualism has to be considered more precisely in order to present solid results (Yairi & Ambrose, 2013).

2.3.5 Natural recovery and persistency

As soon as a child shows symptoms of manifested stuttering, one of the first questions parents are concerned about is about the long-term prospects: is stuttering going to last or might it just vanish as quickly as it first occurred? Most of the children affected, girls especially, will have recovered from stuttering by the time they hit puberty (Natke & Alpermann, 2010). A person who was diagnosed with stuttering in the past is considered recovered if the persons speaks fluently without using any kind of fluency enhancing or mental technique and if stuttering-like disfluencies occur with the same frequency as they do in fluently speaking persons. Recoveries are considered to be permanent if they last for at least 12 months and if a representative sample of spontaneous speech contains less than 3% of syllables stuttered (Yairi & Ambrose, 1999).

A distinction is made between spontaneous (unassisted) and assisted recovery – sometimes also the term remission is used in this context (Guitar, 2014). A recovery is considered as natural if the reduction of disfluencies was not the consequence of a therapeutic intervention. In contrast, an assisted recovery was preceded by a specific therapy or by strategies to enhance fluency by the person alone or with the help of a caregiver (Ingham, Finn, & Bothe, 2005).

Natural recovery, as mentioned above, is most likely to happen by the time a child hits puberty, most commonly taking place within the first four years after onset (Yairi & Ambrose, 2004). Afterwards, chances decline with increasing age and time passed since onset, especially for a natural recovery (Johannsen & Schulze, 2001; Månsson, 2000). During the first two years, and particularly during the first six to twelve months after onset, the rate of natural recovery is highest, indicating that children stuttering for more than two years are at a higher risk of persistent stuttering and therefore also are in a higher need of a therapeutic intervention than children with a shorter time span since onset (Andrews & Harris, 1964; Latterman, 2011). Children with an onset prior to their third birthday have a higher chance for natural recovery than children with an onset after their third birthday (Yairi & Ambrose, 2005). Natural recovery rates, where data was collected retrospectively, vary between 9.5% and 79.1% (Bloodstein & Bernstein Ratner, 2008). While this enormous range is often to be explained by methodological differences, a reliable and comprehensive statement is not possible based on such numbers. It is important in this respect to look at data referring to certain ages and time spans to really get reliable information: before the 10th year of life, rates for natural recovery are about 75% and for children between eight to twelve years around 50% (Johannsen & Schulze, 2001; Månsson, 2000; Yairi & Ambrose, 2005). Boys aged seven or under recover less frequently compared to girls. Natural recoveries, however, are not restricted to childhood years only: they also occur in up to twelve-year-olds and sometimes even in adult years (Anderson & Felsenfeld, 2003; Finn, 2004; Finn, Howard, & Kubala, 2005; Howell, Davis, & Williams, 2008).

The likelihood of recovering from stuttering depends on age, gender and tendency of biological ancestors to recover and is therefore genetically influenced. This hypothesis is supported by the fact that monozygotic twins show higher rates of concordance in stuttering recovery (Dworzynski, Remington, Rijsdijk, Howell, & Plomin, 2007), and also by the fact that the likelihood of a remission is higher if a biological ancestor recovered, too (Yairi & Ambrose, 2005). Unexpectedly, children stuttering mildly in the beginning do not or only marginally differ from severely stuttering peers in terms of their likelihood of recovery (Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; R. V. Watkins, Yairi, & Ambrose, 1999; Wingate, 1976; Yairi & Ambrose, 1999). As a logical consequence, the decision about a potential therapeutic intervention is – at least in the onset stage – not to be made based on stuttering severity. With ongoing stuttering, though, the likelihood of remission seems to be dependent on stuttering severity (Howell & Davis, 2011a).

Also, the relation between general skills in speech and language and the recovery from stuttering is not completely clear, yet. Data on the influence of oral-motor skills, phonological skills, morpho-syntactic skills and lexical skills on the persistency of and recovery from stuttering is inconclusive (Ambrose et al., 2015; Bernstein Ratner, 1997; Howell, 2010; Howell & Davis, 2011a; Paden, Yairi, & Ambrose, 1999; R. V. Watkins, 2005; R. V. Watkins et al., 1999). Whereas in the study by Ambrose et al. (2015) recovered pre-school children and a healthy control group showed better results in general measures of speech and language as well as in lexical tasks than persistently stuttering pre-school children, a German study by Johannsen and Schulze (2001) found out that children recovering within 18 months show a smaller active vocabulary than children recovering later or not at all. Watkins and colleagues (1999) found average and above-average performance on morpho-syntactic and lexical tasks in recovered and persistently stuttering pre-school children. Persistently stuttering children might show developmental disorders concerning their speech-motor skills, their phonological skills and their lexical skills compared to children recovered from stuttering (Nippold, 2018; Smith, Goffman, Sasisekaran, & Weber-Fox, 2012). Since those differences were only found in some children and were, on top of that, very subtle, more research is needed to use language skills as possible predictors for persistent or recovered stuttering. Besides that, other skills, such as rhythmic ones, should be considered. The work in hand collects and discusses information on this topic (see chapter 2.6).

2.4 Assessment, diagnosis, and treatment

After defining stuttering, its symptoms and its epidemiology, the focus of the following section lies on the diagnosis and therapy of stuttering. Since one of the studies in this thesis is concerned with therapy outcome and therapeutic success of interventions, it is of great importance to have the relevant basic information available.

2.4.1 Diagnosing stuttering

The diagnosis of stuttering aims at assessing onset, duration and extent of the disorders as well as its impact on social functionality, activity and participation. Therefore, it aims to consider the quality of life of a person who stutters, to detect comorbidities, to evaluate the need for a therapeutic intervention and, if so, to determine the adequate form of treatment and its possible outcome (Guitar, 2014; Van Riper, 1982).

A structural framework for diagnosis, treatment and monitoring of treatment effectivity is provided in form of the *International Classification of Functioning, Disability and Health (ICF)* by the World Health Organization (2001), offering an internationally uniform and standardized language for the description of the functional health status of an individual in terms of his or her physical and mental ability and disability as an interaction of the physical functions, structures, the social impairment and the personal context as well as factors within the individual’s environment (WHO, 2001). The classification of stuttering into this framework has been done previously and is depicted in figure 2.1 (Yaruss, 2007; Yaruss & Quesal, 2004).

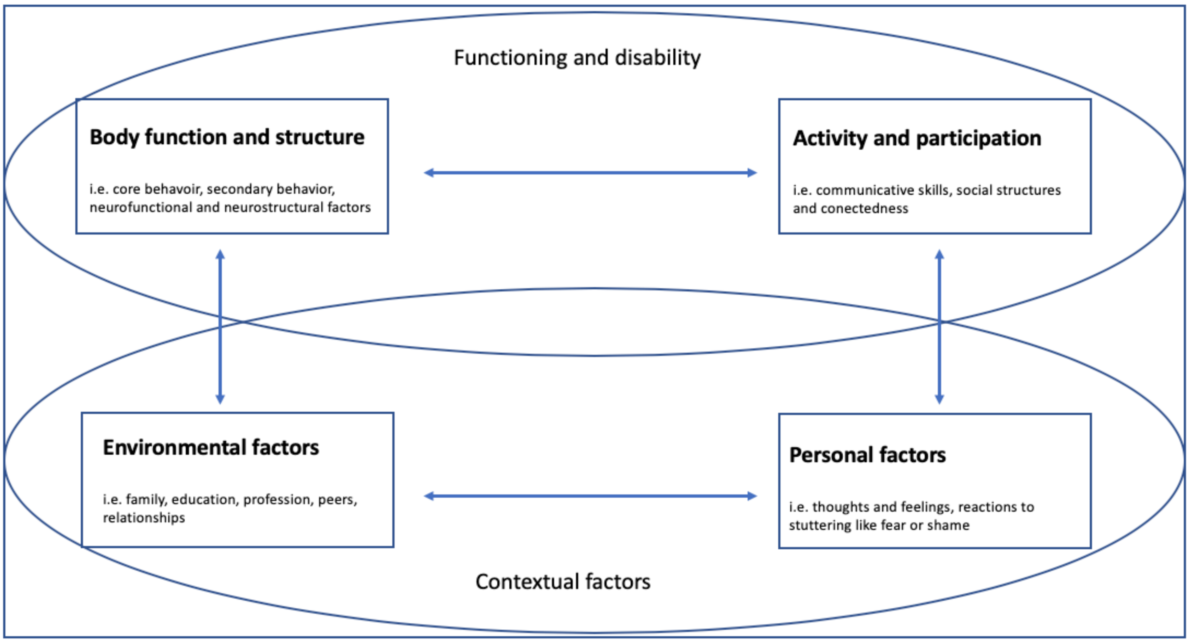


Figure 2.1 Stuttering in the context of the International Classification of Functioning, Disability and Health in a modification of Cook (2013)

Prior to the diagnosis of stuttering, it is quite common to do a screening and a detailed interview about the medical history – in case of younger children mostly with parents, in case of adolescents or adults with the client him- or herself (Guitar, 2014) to get a first impression on factors like variability in stuttering and the impact of the disorder on the client's life in family, school/professional life, relationships and further *environmental factors*. Also, it is important to get an impression about the individual's thoughts and feelings towards his or her stuttering and further *personal factors*. Taking the model of the ICF to evaluate the need for therapy, this need is firstly determined by the core symptoms, since one of the primary goals of a treatment for stuttering is the increase in fluency, as depicted in *body function and structure*. Furthermore, an unrestricted participation through the depletion of psychosocial fears in the context of communication (see *Activity and participation*) is one of the major goals. A professional and comprehensive diagnosis of stuttering according to the ICF is thus based on a medical history, with an assessment of core symptoms and secondary behavior, the psychosocial impact, skills in communication and social behavior as well as reactions from the social environment. The assessment of stuttering is composed of objective (quantity and quality of core symptoms and motor behavior), descriptive (description of stuttering symptoms through a third person) as well as self-perceived (impact of stuttering on the daily life) measures (WHO, 2001). Two of the most used methods of assessment are described in the following.

One of the most valid methods internationally used to quantify stuttering severity– also used in the thesis at hand– is the Stuttering Severity Instrument – Fourth edition (Riley, 2009). It is designed for children from 2 years, adolescents and adults and captures three aspects of stuttering: the frequency of stuttering symptoms (percentage of syllables stuttered) in spontaneous speech and, if the person is already alphabetized, also in a reading excerpt; the mean duration of the three longest symptoms and non-speech behavior such as grimaces, head, or limb movement are rated on scales by a professional. Those raw values allow a classification into five different degrees of stuttering severity ranging from very mild to very severe. It therefore is an objective instrument to assess stuttering severity. There is no German standardization, but since the test is not bound to one language in its applicability, it is also appropriate to use the German translation with American norm values (Sandrieser & Schneider, 2008). For the study in the paper at hand, the Stuttering Severity Instrument in the third edition (SSI-3; Riley, 1994) was used, as therapists of the courses had decided to use this version. However, SSI-3 and SSI-4 only differ in terms of statistic criteria and normative data but not in implementation or content (Riley, 1994, 2009; Sandrieser & Schneider, 2008).

Since the SSI-4 is an objective measure to assess severity but not the subjective impact of stuttering on an individual's life, it is important in the context of ICF to add an instrument accounting for those subjective measures of life quality and functioning in everyday life. One of the most commonly used instruments, which was again used for the study of the thesis at hand, is a questionnaire named OASES (Overall Assessment of the Speakers' Experience with Stuttering, Yaruss & Quesal, 2006, 2008). It was developed based on the ICF and has been accessible in its German translation since 2016. The OASES exists in three forms, one for schoolchildren (aged 7-12 years), one for teenagers (aged 13-17 years) and one for adults (from 18 years). All three forms of the OASES are long and markedly comprehensive. Every item of the OASES is answered on a five-point rating scale; calculations can be obtained for single subtests or for the whole questionnaire. Raw values can be classified ranging from mild to severe. It therefore is a very sensitive instrument to capture the impact of stuttering in situations of daily life and also quantifies changes induced by therapeutic interventions (Yaruss & Quesal, 2006, 2008).

2.4.2 Differential diagnosis

After discussing the assessment and diagnosis of stuttering, it is important to also mention frequent differential diagnoses, that must be clearly distinguished from stuttering.

2.4.2.1 Normal disfluency

This differential diagnosis is a very important one since it is the basis of any further therapeutic intervention. At the same time, this decision between normal disfluency and manifested stuttering is often quite difficult (Guitar, 2014). During speech development, especially at the age from two and a half to four years, nearly all children display disfluencies (Starkweather, 1987). Those normal and age-appropriate disfluencies often occur in form of repetitions of words or phrases and are usually not accompanied by signs of exhaustion. Furthermore, revisions, termination of sentences, short pauses or tension-free prolongations of up to one second are considered symptoms of normal disfluency. Interjections and pauses, in particular, have mostly a clear origin in speech planning rather than in stuttering-like disfluencies. Furthermore they do not influence rhythm or prosody (Guitar, 2014; Natke & Alpermann, 2010; Ochsenkühn et al., 2015). It is believed that those kinds of disfluencies originate from an immature speech system, since children at this age are still in the process of coordinating and developing individual achievements within the speech system. Articulation, lexical retrieval and syntactic planning are an enormous accomplishment. On top of that, there is evidence for the occurrence of those disfluencies especially within those stages of speech and language development where more complex structures are learned and used for the first time (Bloodstein & Bernstein Ratner, 2008; Zackheim & Conture, 2003). Usually, those overextensions of the speech system do not last longer than six months (Ochsenkühn et al., 2015). Investigating normal disfluencies in fluently speaking children, Guitar (2014) observed a mean value for disfluencies of seven out of 100 words, with repetitions being the most frequent ones. Even adults show up to 20% of disfluencies without being categorized as PWS. Those kinds of disfluencies are mostly used functionally for further speech planning (Starkweather, 1987).

Early stuttering, however, shows qualitative differences often in very early stages even if it is not possible to classify those kinds of disfluencies yet: symptoms of normal disfluencies as well as real stuttering symptoms can be observed (Ochsenkühn et al., 2015). For the purpose of differential diagnosis, it is not recommended to merely consider the duration of those symptoms, since some children tend to remain in this intermediate state for some time. Beginning stuttering can contain all symptoms of normal disfluencies but only few of manifested stuttering at the same time: especially repetitions of sounds with an insertion of the schwa sound as well as the occurrence of a glottis-stop is a sign for beginning stuttering, since those symptoms represent the change of a word in its form. Additionally, speech tempo and speech rhythm tend to deviate significantly from the normal range in regard to repetitions of syllables and sounds. A further signal for beginning stuttering is the rise of effort during speech production: In prolongations and blocks significantly more tension of articulatory muscles is observable (Bernstein Ratner, 1997; Bosshardt, 2010; Guitar, 2014; Reilly et al., 2009). As mentioned before, the line between normal disfluencies and beginning stuttering is drawn at a disfluency level of three percent (Natke & Alpermann, 2010; Ochsenkühn et al., 2015). Normal disfluencies are furthermore ruled out if a child already shows secondary behavior such as head or limb movement, escape behavior or avoidance behavior. In case of an elevated risk for persistent stuttering (i.e. family history of stuttering, male gender etc.), an early intervention is recommended (Reilly, Onslow, & Packman, 2013).

In manifested stuttering, all symptoms of normal disfluencies can appear. They do, however, appear more intense concerning core symptoms and secondary behavior as well as in the awareness of the disorder. Frequently, this leads to a severe level of suffering. A commonly used guideline for pediatricians and professionals was developed to have a clear scheme for differentiating between normal disfluency and stuttering. According to this guideline a child is at the stage of manifested stuttering if one or more of the following six conditions is met: the *duration* of symptoms has been over six months, tensions, tremors or associated movements of head or limb have occurred during the *process* of disfluencies, the *symptoms are accompanied* by a rise in tension, pitch level or volume and a visible exhaustion, the *reaction of the child* towards his disfluencies are clearly recognizable (it reacts in form of avoidance behavior or escape behavior), *parents are concerned* and report a manifestation and a *family predisposition* for stuttering is reported (Johannsen & Schulze, 1992, 2008). The differentiation between normal disfluency, beginning and manifested stuttering is especially relevant for further prevention or therapy. Certain critical signals, as mentioned above, should therefore be considered in this decision.

2.4.2.2 Other fluency disorders

Stuttering is not only to be differentiated from normal disfluency. Other disorders of speech fluency must also be taken into consideration when defining stuttering. Other fluency disorders, such as cluttering, neurogenic or psychogenic stuttering, are to be distinguished from developmental stuttering (also: idiopathic stuttering), which is the center of the thesis at hand. For reasons of readability, the term stuttering is used synonymously for developmental/idiopathic stuttering. In the following, those diagnoses similar to stuttering are described with a focus on the distinction to idiopathic or developmental stuttering. Speech fluency is defined by aspects concerning the continuity, rate and effort with which speech is being produced (American-Speech-Language-Hearing-Association, 1999). Pathological aberrations of the normal speech flow are mostly caused by stuttering or cluttering.

Cluttering is sometimes quite similar to stuttering. By definition, cluttering is a speech disorder which is characterized by a high and unregular speech rate. Furthermore, one of the four following symptoms has to be observed: a reduced intelligibility caused by phonetic or phonological abnormality, a changed prosody, an unclear articulation and an above-average number of disfluencies (Myers, Bakker, St.Louis, & Raphael, 2012). It is quite common that cluttering and stuttering co-occur in an individual, making differential diagnosis difficult (Howell & Davis, 2011b; Van Zaalen-op't Hof, Wijnen, & De Jonckere, 2009). One of the most prominent features of cluttering is the very rapid and unclear articulation. It causes repetitions, elisions and fusions of words, syllables, and sounds. Intelligibility is affected negatively by further reductions of consonantal clusters or eliminations of suffixes. At the same time, the awareness of the disorder is much lower in cluttering and symptoms can be moderated significantly by focusing on a lower speech rate (Myers et al., 2012; Van Zaalen-op't Hof et al., 2009).

As already mentioned above, further categories of stuttering exist besides developmental stuttering. These are explained briefly in the following paragraphs and visualized in figure 2.2 (Neumann et al., 2017): acquired stuttering which can be sub-divided into neurogenic stuttering and psychogenic stuttering, and originary syndromal stuttering, which is in contrast to originary non-syndromal stuttering clearly linked to a genetic syndrome.

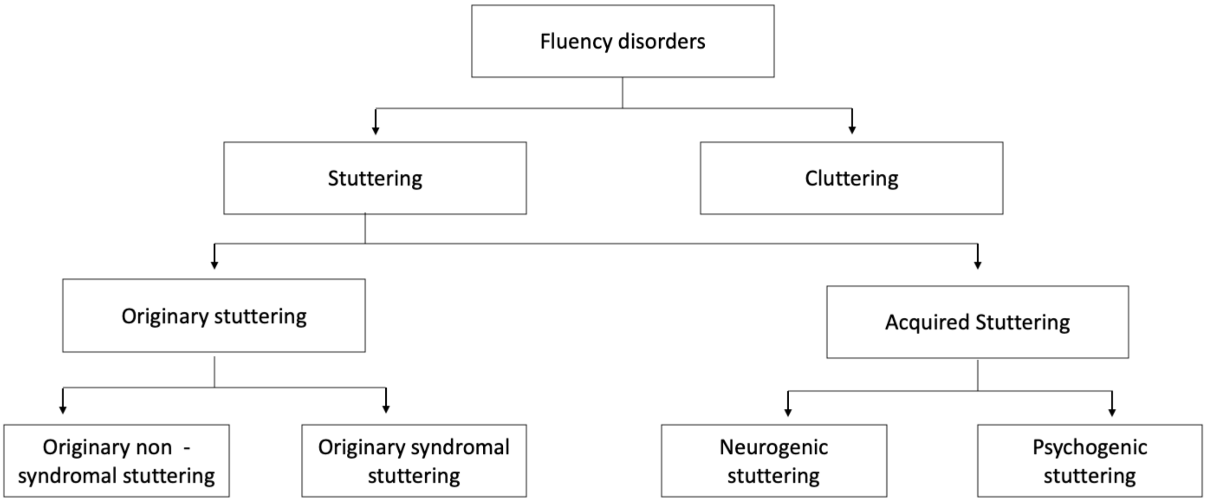


Figure 2.2 Classification of different fluency disorders: adapted figure from Neumann et al. (2017)

Neurogenic stuttering, which has a sudden onset at any age but is more likely in adults, is caused by a definable brain damage through events like a stroke or a head trauma (Sommer & Büchel, 2004). Neuropathological correlates of neurogenic stuttering can be quite distinct and are not restricted to a certain cerebral or cerebellar area. However, they are most frequent after lesions in subcortical structures such as the basal ganglia (BG) and the pons (Lundgren, Helm-Estabrooks, & Klein, 2010). Furthermore, lesions of the thalamus can result in neurogenic stuttering (Van Borsel, Van Der Made, & Santens, 2003). It has also been reported that some psychotropic drugs or alcohol abuse can lead to similar symptoms (Boyd, Dworzynski, & Howell, 2011; Grover, Verma, & Nebhinani, 2012). It is quite common for neurogenic stuttering to co-occur with other neurogenic speech and language disorders such as aphasia or dysarthria, which makes a clear differential diagnosis even more difficult (Bosshardt, 2008). In this kind of stuttering, secondary behavior often is not as pronounced as in developmental stuttering. Additionally, core symptoms are less variable and not influenced by external factors (i.e. stressful communicative situation, fear etc.). Therefore, neurogenic stuttering merely shows fluctuations in severity and there is neither an influence of the word-class nor of specific syllables or sounds (Lundgren et al., 2010; Ochsenkühn et al., 2015).

An even more rare form of acquired stuttering is psychogenic stuttering. This type of stuttering with a sudden onset, usually in adulthood, is clearly associated with a psychiatric disorder or a psychological trauma. It is most frequently associated with an anxiety disorder or depression, but schizophrenia and post-traumatic stress disorders can also cause psychogenic stuttering (Mahr & Leith, 1992; Sandrieser & Schneider, 2008). The exact cause of outbreak is still not completely understood. It is furthermore unclear whether there is a link between developmental stuttering and acquired stuttering. The pattern in symptoms can be used for differential diagnostics. While symptoms are usually to be found in the beginning of the word in developmental stuttering, symptoms in acquired stuttering occur in any word position and are not influenced by external factors or stressors (Bosshardt, 2008; Lundgren et al., 2010; Mahr & Leith, 1992).

Originary syndromal stuttering is caused by distinct genetic syndromes, that are often associated with stuttering or stuttering-like syndromes. Stuttering is listed as a feature of various syndromes, in particular of Down Syndrome, Prader-Willi-Syndrome, Fragile-X-Syndrome, Turner-Syndrome and Tourette-Syndrome. An overview of those syndromes states that persons affected show disfluencies, of which not all can be assigned to stuttering (Van Borsel & Tetnowski, 2007). Many of those syndromes are characterized by different grades of intellectual disability leading to a higher prevalence of stuttering compared to the non-affected population, since syndromes associated with intellectual disability frequently show disfluencies, which are not always stuttering-like (Van Borsel & Tetnowski, 2007).

2.4.3 Co-occurring disorders

While stuttering is not uncommonly associated with further disorders, it is not clear to what extent those further disorders are causes or consequences of stuttering or whether they are simply co-existent. In literature, reported numbers on co-occurring disorders vary significantly due to different methods and divergent criteria for diagnoses. However, it is very important to clarify which disorders are to be clearly separated from stuttering, since they must not co-occur with stuttering by definition, and those that frequently co-occur with stuttering (von Tiling et al., 2014). For the present thesis, this knowledge is relevant with respect to the participants of all three studies, since they were compiled with specific regard to co-occurring disorders: to ascribe differences in their performance to stuttering, children and adolescents with co-occurring disorders were excluded from the studies.

Co-occurring disorders of stuttering were investigated by comparing stuttering and nonstuttering persons with acknowledged data on prevalence. Furthermore, surveys with speech therapists yielded information for the cumulated occurrence of symptoms of specific language impairment and of learning- and reading disorders (Arndt & Healey, 2001): according to those surveys of 241 American speech-language pathologists, 44% of children affected by stuttering aged three to twenty showed additional disorders (i.e. phonology disorders, articulation disorders, other speech or language disorders). Another study by Blood and colleagues (2003) consulted 1,242 American speech-language pathologists in a mail survey with clear results: according to the specialists, 62.8 % of stuttering children had further co-occurring disorders in speech or language or other disorders (i.e. learning disabilities, literacy disorders or attention deficit disorders). Furthermore, researchers found out that significantly more boys were affected by a co-occurring disorder than girls. If a co-occurring disorder in speech or language is present as early as in pre-school age, it is usually seen as an indicator for persistent stuttering (Paden et al., 1999). However, it is still doubtful whether the occurance of stuttering and general language skills are linked, newer data does not tend to support this idea (Nippold, 2012, 2018).

When stuttering is already persistent in adolescent or adults, symptoms of an anxiety disorder accompany it. Sometimes those symptoms are as severe as in socially phobic patients. Stuttering and social anxieties are linked so closely that stuttering is an exclusion criterion for the diagnosis of a social anxiety disorder (Bosshardt, 2008).

In summary it can be stated that in numerous children a co-occurring disorder of speech or language is present, most frequent are disorders in articulation or phonology. In adolescents or adults, however, anxieties are most likely to occur in combination with persistent stuttering, often gaining a very strong and burdensome extent during the process of chronification (Bosshardt, 2008).

2.4.4 Treatment

At present, there are various therapies, programs and interventions aiming to help PWS regain control over their way of speaking, increase fluency or cope with secondary behaviors and stuttering-related fears. Those different approaches as well as certain criteria that must be met for an evidence-based therapy program and the goals of specific therapies will be explained in the following, with a specific focus being put on the therapeutic approach used on stuttering participants in the present study.

2.4.4.1 Treatment criteria and goals

Persons who decide to undergo treatment have the right to get the best treatment currently available and evaluated scientifically. In the field of treating stuttering the diversity is enormous, since there have been lots of different theories about curing stuttering in the history of fluency disorders. Furthermore, it has not been mandatory until now to prove that the chosen treatment was actually evidence-based (Guitar, 2014). Randomized-controlled studies are necessary for the development and validation of evidence-based treatments and although that is very complex, effortful and also difficult to do in the field of stuttering due to factors like waiting-control groups or sufficiently big intervention groups, there has been much progress during the last years (Bloodstein & Bernstein Ratner, 2008; Guitar, 2014; Neumann et al., 2017).

The effect of stuttering treatment can be described by three measures of success: most frequent is the report of *efficacy* in controlled clinical settings. More important, but a lot harder to find in literature, are reports of *effectiveness*, concerning the impact of a treatment on every-day life. *Efficiency*, the relation of therapeutic effect and therapeutic effort is also an economically relevant measure of success in the treatment of stuttering (Conture, 1996; H.A. Euler, Lange, Schroeder, & Neumann, 2014; Guitar, 2014). Furthermore, Bloodstein and Bernstein Ratner (2008) specified twelve criteria conserning research methods that have to be met in order to proclaim a treatment as effective and successful. Those criteria help judge a treatment and make the comparison of different interventions more transparent. For the thesis at hand, a comprehensive description of different concepts and interventions is not possible or reasonable, which is why the descriptions will be limited to the two groups of established therapeutic interventions. They are also relevant for the work at hand, since the group of PWS tested for the three studies of this thesis was treated with a method-combination of those two interventions.

Basically, the treatment of stuttering has two major goals: On the one hand, disfluencies should be reduced and fluency should be promoted. On the other hand, a reduction of the emotional burden associated with stuttering as well as of the negative cognitive reactions and social fears is just as important as the treatment of the disfluency itself (von Tiling et al., 2014). The individual focus is always to be determined with the patient him- or herself. Since the actual causes of stuttering still remain unknown, there is no causal therapy that is equally successful in all persons affected (Natke & Alpermann, 2010).

Whereas in young children the main focus mostly lies on the recovery from stuttering, in adolescents and adults it is mostly the handling of disfluencies and the reduction of secondary behavior that is central in treating stuttering (Guitar, 2014). During the treatment of childhood stuttering, it is quite common to use indirect approaches that aim to influence factors triggering and maintaining stuttering. Therefore, a distinction between direct, indirect and operant approaches is made, especially regarding the therapy of children and adolescents. While indirect approaches like the Palin PCI approach (Kelman & Nicholas, 2008) mainly focus on advising and training parents and strengthening the child's self-confidence, direct approaches work on speech and disfluencies directly, mostly by introducing certain techniques (Sandrieser & Schneider, 2008). Operant approaches, however, work on enhancing fluency, like the Lidcombe program (Latterman, 2008; Onslow, Packman, & Harrison, 2003). Direct approaches can again be divided into two main directions, which are methods of speech restructuring, of which fluency shaping is the most commonly known, and modification approaches as well as combined approaches uniting techniques of both directions (Blomgren, 2013; Guitar, 2014). As mentioned above, a restriction to the description of the two most common approaches is reasonable for the paper at hand.

2.4.4.2 Fluency shaping

Fluency shaping is the best-known form of speech restructuring and is considered a global technique since it aims at changing the whole process of articulation. It is supposed to be a systematic construction of a fluent way of speaking, teaching PWS a new way of articulating (Natke & Alpermann, 2010). Goldiamond and Webster are considered to be precursors of this technique (Goldiamond, 1965; R. Webster, 1977). Prolonged Speech is considered to be the most popular and empirically best researched global technique: PWS are trained to acquire a completely new way of speaking, which prevents stuttering symptoms from occuring by applying a decelerated mode of speaking, with soft and smooth voicing and a continuous breathing (von Tiling et al., 2014; R. Webster, 1977). With lots of practice in a highly structured manner, this initially unnatural and monotone way of speaking can become more and more natural and can be applied in situations of every-daily life (Guitar, 2014; Natke & Alpermann, 2010). First, people who stutter learn this technique at very slow pace and at a low level of demands, later the application of the technique is increased in demands until the level of daily speaking is achieved. The training of this global technique is often done with a software specifically developed for this purpose (Guitar, 2014). Since methods of speech restructuring assume that stuttering is not likely to occur when applying the techniques, most of them do not address feelings and attitudes towards stuttering.

However, learning this new way of speaking requires an intense practice phase, which is why most of those programs are held as an intensive therapy course in a clinical setting. Examples of well-researched and evidence-based therapy programs offering fluency shaping are the *Camperdown Program* (O'Brian, Cream, Packman, & Onslow, 2001; O'Brian, Onslow, Cream, & Packman, 2003) or the Kasseler Stottertherapie (H. A. Euler, Gudenberg, v. Jung, & Neumann, 2009). A recent study demonstrated that those techniques can also be learned successfully and effectively in an outpatient setting (O'Brian et al., 2003).

2.4.4.3 Stuttering modification

Local techniques that are only used on the specific sounds or syllables where symptoms are being expected are summarized under the term *modification*. Words which are expected to be spoken without any symptoms are produced normally and without any changes induced by speech techniques (von Tiling et al., 2014). This method reflects a completely different way of treating stuttering and goes back to Charles Van Riper (1971). At the time, this method was a very new and innovative way of treating stuttering. It assumed that a vast amount of the conspicuous behavior in stuttering is derived from learned reactions to interruptions of the onward flow of speech. The main goal of this method, therefore, was to reduce fear and shame by teaching PWS to change their stuttering in a way that is less disruptive and more controllable in communication (Van Riper, 1971). Thus, modification focuses on other contents than fluency shaping: not the way of speaking but rather the factors contributing to negative emotions around the core symptoms are in the center of the therapeutic intervention (Natke & Alpermann, 2010; Zückner, 2008). Modification is also known as a non-avoidance method, since one of its main goals is the reduction of avoidance and escape behavior as well as the reduction of frustration and fear. However, this method also contains techniques to make stuttering symptoms controllable. This approach for the treatment of mostly adolescents and adults consists of a four-phase model to be implemented in stuttering treatment (Guitar, 2014; Van Riper, 1971, 1982; van Riper & Emerick, 1984).

During the first phase, which is the so-called *identification,* a detailed analysis of core behavior and secondary symptoms is at the center of the therapeutic intervention. Feelings and attitudes towards stuttering but also the self-perception of core symptoms are essential to be able to change those in later steps in the process of modification. This identification is often done with the help of video sequences by confronting the person affected with his or her specific symptoms. This phase of identification is followed by *desensitization* against the loss of control during symptoms and reactions of listeners. Emotions are analyzed and modified with cognitive-therapeutic interventions to reduce avoidance and escape behavior leading to a more relaxed dealing with one's own symptoms. In the following phase of *modification* local techniques are practiced controlling stuttering symptoms when they appear. Techniques can be applied either during a symptom to reduce tension or duration, or prior to a symptom, to avoid an anticipated symptom. However, to correctly use those techniques, PWS must have abandoned their avoidance behavior and escape behavior, thus permitting core symptoms to occur. One of the most popular techniques is the so-called *Pull-Out,* which is used directly at the moment of stuttering: the symptom has to be stopped (often also described as freezing the symptom) and the situation is afterwards resolved by a slow and relaxed way of continuing the phonation (Natke & Alpermann, 2010; Van Riper, 1982; van Riper & Emerick, 1984). In the long term, this continued handling of disfluent parts leads to a reduction in the rate of stuttering symptoms and to a feeling of control over one's speaking. Fluent parts of the articulation are not changed in any way. While this method does not promise a complete reduction of stuttering symptoms, the feeling of regained control over speech fluency further promotes the person's confidence, leading to a higher degree of speech fluency and to a reduction of fear. All modification therapies require an implementation in every-day life. A therapeutic support to get accustomed to the application of the newly learned techniques beyond the therapeutic setting is given in the phase of *stabilization*. Furthermore, the prevention of relapses is an important issue during aftercare (Van Riper, 1982; van Riper & Emerick, 1984). Stuttering modification is applied in the outpatient as well as in the clinical setting and while some therapies have undergone evaluation, evidence is weaker than in fluency shaping methods (Natke, Alpermann, Heil, Kuckenberg, & Zückner, 2010; Zückner, 2008). Techniques of modification therapies are usually easier to learn and to apply long-term, since they allow a spontaneous way of speaking and they also include working on feelings and fears towards stuttering. However, in applying local techniques, the control over one's speaking is more limited than in applying the technique of fluency shaping and the experience of losing control is more likely to occur when using modification techniques (von Tiling et al., 2014).

2.4.4.4 Combination of fluency shaping and modification

Both methods have their advantages and disadvantages. Therefore, the combination of fluency shaping and modification has been established in the therapeutic field and seems reasonable (Guitar, 2014; Natke & Alpermann, 2010). However, evidence for those combined therapeutic approaches is still weak (Blomgren, 2013; Langevin et al., 2006; Langevin, Kully, Teshima, Hagler, & Narasimha Prasad, 2010; Metten, Zückner, & Rosenberger, 2007). In some interventions, patients undergo a determined and structured program, in others therapeutic goals and elements are deduced from the individual symptoms and requirements of the patient (Guitar, 2014).

Authors and therapists favoring a combination of methods assume that PWS can frequently reach fluent periods but show a higher rate of symptoms in stressful situations. Thus, it is their therapeutic goal to give patients a choice in those situations: If fluent speech is desired in one situation, the patient can choose to use techniques of fluency shaping. If a patient wants to speak rather fluently but does not have the capacities to switch to a complete usage of fluency shaping techniques, he or she can also reach a high level of fluency by using local techniques of modification. Finally, there is the option of tolerable stuttering without fear or secondary symptoms, if a patient is not able or motivated to use further fluency enhancing techniques. Experts supporting a combination of methods hence want to achieve a maximum of flexibility, individuality and the possibility to decide freely in each situation (Guitar, 2014; Natke & Alpermann, 2010). Internationally, this trend towards integrative therapeutic concepts is also growing, sometimes even adding elements of cognitive-therapeutic interventions (Langevin et al., 2006). To date, there is no evidence for the superiority of a method-combined approach, but there are hints that a method-combined intervention shows similar results to fluency shaping or modification (H.A. Euler et al., 2014). Participants of the studies conducted for the paper at hand were treated following the program of a method-combined intervention (Thum & Mayer, 2014).

2.5 What causes stuttering?

There are not only a huge number of different therapeutic approaches to stuttering but also many theories about the origin of stuttering that have been postulated throughout the centuries:

first written records are found in the Bible, where Moses was considered a stutterer (von Tiling et al., 2014), and in ancient Greece, where the dryness of the tongue was held responsible for the stuttering phenomenon (Sommer & Büchel, 2004). This led to a mostly surgical orientation in treatment, causing further lesions or disabilities. During the 20th century, stuttering was considered of psychogenic origin and therefore treated with psychoanalytical approaches or methods of behavioral therapy. However, studies investigating parameters like personality traits or parent-child interactions could not find systematic psychological patterns linked to stuttering (Andrews et al., 1983).

Theories on the causes and origins of stuttering have to explain a variety of phenomena that seem enigmatic at first sight: why is there such a broad spectrum of variability? The same person is able to speak completely fluently in one situation but struggles to articulate one fluent word in another situation. This phenomenon hints towards the fact that there is a psychological effect behind it. Why do most of the children affected by stuttering recover from it (naturally) after a short period and others do not? This fact, however, suggests a genetic component. And finally, there also has to be a sufficient explanation for the broad range of symptoms, their severity and frequency, which leads to extreme variability in interindividual symptoms. Based on this complex phenomenology, nowadays not monocausal but multicausal models for the emergence and persistence of stuttering are assumed. They all have in common that stuttering is seen as a phenomenon of individually differing conditions and an interaction of many factors contributing to it. These observations have resulted in a two-factor model in the development of stuttering: whereas the first factor is considered to be the actual cause of the disorder in form of abnormalities in the function or structure of the central nervous system, the second factor maintains or enhances the first one by avoidance learning (Sommer & Büchel, 2004). One has to be very cautions, however, in calling the latter factor psychological or psychogenic, since neuroscience has demonstrated that learning also leads to changes in the brain that are actually measurable (Kandel & O'Dell, 1992). In the following chapters, those factors regarding the causes of stuttering will be explained.

2.5.1 Genetic and environmental factors

With behavioral genetic research (i.e. twin studies, family studies, adoption studies) it is possible to evaluate the genetic component and the influence of environmental factors contributing to the variance of characteristic attributes, whereas molecular genetic research is able to localize specific genes, leading to individually different characteristics of a phenotypical attribute (Dworzynski et al., 2007; Frigerio-Domingues & Drayna, 2017; Kraft & Yairi, 2012).

Behavioral genetic research confirmed a high heritability in stuttering (Andrews, Morris-Yates, Howie, & Martin, 1991; Dworzynski et al., 2007; Felsenfeld et al., 2000; Frigerio-Domingues & Drayna, 2017; Kraft & Yairi, 2012; Ooki, 2005). Monozygotic twins show a significantly higher rate of concordance regarding the probable occurrence of stuttering than dizygotic twins of the same sex, namely 44.9% in the first case compared to 12.0% in the latter case. This value was calculated as a mean of six studies and weighted according to sample size (Andrews et al., 1991; Dworzynski et al., 2007; Felsenfeld et al., 2000; Godai, Tatarelli, & Bonanni, 1976; Howie, 1981a; Neumann et al., 2016). In adult PWS, heritability lies between 70% (Felsenfeld et al., 2000) and 80% (Fagnani, Fibiger, Skytthe, & Hjelmorg, 2011; Ooki, 2005; Rautakoski, Hannus, Simberg, Sandnabba, & Santtila, 2012), whereas in children who stutter (CWS), heritability is considered to be around 60% and is therefore a little lower (Dworzynski et al., 2007). It is now scientifically proven for children that not only the emergence of but also natural recovery from stuttering is inherited (Ambrose, Cox, & Yairi, 1997; Dworzynski et al., 2007). However, those findings only apply to the probability of occurrence and not to the severity of stuttering itself (Howie, 1981b).

Heritability is a concept for populations and does not allow a specific prediction on the genetic influence for an individual, but it does allow a prediction on the probability of stuttering if a biological relative is also affected by stuttering. The occurrence of stuttering in biological relatives predisposes towards the emergence of stuttering and increases the risk from 5% (in an unselected male population) to about 20% in populations with male relatives affected by stuttering. The risk is even higher for sons of stuttering women while the familial clustering is considerably lower between female relatives (Kidd, 1980, 1981; Yairi & Ambrose, 2005).

The remaining variance of 30% or less stems from environmental factors: behavioral genetic research differentiates between environmental impacts affecting siblings equally and making them more similar in comparison to children of other families (i.e. shared environment) and other environmental impacts (i.e. non-shared environment). Results of behavioral genetic research mostly agree that etiologically relevant environmental impacts on the emergence of stuttering derive from the category of *non-shared environment* (Andrews et al., 1991; Fagnani et al., 2011; Felsenfeld et al., 2000; Ooki, 2005). Hence, it is unlikely that a child’s language environment is a relevant factor in the origin of stuttering. While the educational style does not seem to be responsible for the emergence of stuttering, insecure parents without adequate counselling might show an unfavorable way of interacting with the affected child (i.e. exhortations like “Think before speaking” etc.) leading to possible manifestations of stuttering severity or psychological consequences for the child (i.e. insecurity, loss of communicative naturalness).

The amount of variance of the non-shared environment cannot be derived from behavioral genetic results. This is because an environment consists not only of all things that are consciously perceived by a person but also influences of all kinds of environment, like social environment, uterine environment, physiological-anatomical environment and the intellectual world. Furthermore, coincidence also plays a role in non-shared environment and development is a complex field where genes start off a network of interactions with molecular, cellular, physiological, behavioral, and social components. Therefore, it is barely possible to say what the specific causes for the emergence of stuttering are in a non-shared environment. A retrospective analysis of causes and the interpretation of social or psychological events that preceded the onset of stuttering (i.e. traumatic experience) are typically mistakes in attribution, equating coincidence with causality (Fagnani et al., 2011; Felsenfeld et al., 2000; Rautakoski et al., 2012).

Molecular genetic research has found over a dozen loci for stuttering on chromosomes 1, 2, 3, 5, 7, 9, 12, 13, 15, 16, 18 and 21 (Kang et al., 2010; Kraft & Yairi, 2012; Shugart et al., 2004; Suresh et al., 2006; Wittke-Thompson et al., 2007). Furthermore, a disposition location on chromosome eight was identified, sharing alleles[1] with all persons who do not stutter (PWNS) and which was therefore declared a protective factor (Kraft, Below, Huff, & Yu, 2015). According to present knowledge, stuttering is considered a multifactorial, polygenetic disorder with different loci and genome-environment-interactions. It is not completely clear yet to what extent they lead to a first onset or persistent stuttering. Regions associated with stuttering in former and persistently stuttering persons were found on chromosome 9, and in persistent stuttering in chromosome 15. A linkage between stuttering and chromosome 9 was only found in men, the linkage in women was in chromosome 21 (Suresh et al., 2006). The effects seem to be largely of an additive nature, indicating a risk-threshold model: according to such a model, the risk of developing persistent stuttering is distributed evenly and rises with the number of loci affected. When a certain threshold is exceeded, stuttering would occur. According to this model, the critical threshold would be higher in girls than in boys (Dworzynski et al., 2007).

Genetic evidence leads to consequences for the assessment, prognosis, treatment and counselling of PWS. Heritability does not exclude variability: effective therapeutic interventions can raise the phenotypical differences regarding stuttering, leading to a reduction

---

[1] Allels are different variations of a gene at a certain gene locus, which is relevant for the expression of specific characteristics (Frigerio-Domingues & Drayna, 2017)

in the variance that is attributable to genetic variations and heritability decreases. Therefore, a high risk of hereditary stuttering does not affect the benefit of a symptom-oriented therapeutic intervention (Howie, 1981a; Yairi & Ambrose, 2005). With the robust results concerning the environmental influences on the onset of stuttering all being from the non-shared environment, major consequences for the counselling of parents arise: besides hereditary factors, parents are not to be blamed for the primary onset of stuttering in their children and are, therefore, not to be held responsible.

2.5.2 Neurological factors

Neuroimaging techniques have provided new information on the origins of stuttering during the last years. It has been known for quite some time that brain damage can cause neurogenic stuttering. However, it was new techniques, such as voxel-based morphometry - an objective technique allowing to estimate the amount of specific tissue in a specific neurologic location - that proved that neurological abnormalities also exist in developmental stuttering (Beal, Gracco, Brettschneider, Kroll, & De Nil, 2013; Lundgren et al., 2010). Thanks to neuroimaging techniques it was possible to analyze structural and functional correlates of stuttering by comparing patterns of fluently speaking persons with those of PWS. Results proved differences concerning the activation as well as the structure of areas relevant for speech perception and production (Sommer & Büchel, 2004). At the present moment, there are many studies on neurological evidence in adult PWS, but significantly fewer were conducted with children and adolescents who stutter due to the fact that some methods are not appropriate for children or allowed within this age group (Howell, 2011). Recent studies also tried to account for this fact, since this younger age group represents a very interesting population regarding the question of originary neurologic aberrations or adaptive processes. However, it is hardly possible to present all results on this topic in their completeness. Therefore, the following two chapters shall focus on the most relevant and recent information.

2.5.2.1 Neuromorphological evidence

Changes in density of gray matter, in cortical thickness, in gyration, in the integrity of white fiber tracts and in the structural connectivity can hint towards pathological or adaptive processes, with a minor expression usually indicating a primary pathological incident and a stronger expression of matter indicating adaptive changes (May & Gaser, 2006). Therefore, a reduction in gray and white matter in PWS would point to loci of genetic origin, whereas an increase in matter would rather be a sign of compensatory processes (Kell et al., 2009; Neumann

& Euler, 2010). Most studies make use of different techniques to measure changes in gray or white matter to draw conclusions on the integrity of certain fiber tracts or areas. For a better understanding, results are presented regarding the neuromorphologic subcategory investigated. With magnetic resonance imaging (MRI), differences concerning the symmetry and structural differences in adults PWS were found: the planum temporale (PT), an area posterior to the auditory cortex, strongly lateralized and relevant for speech, was demonstrated to be significantly bigger in adult PWS compared to fluently speaking persons. Consequently, a reduction of the usual asymmetry between right and left hemisphere in the PT and in auditory regions was observed (Foundas, Bollich, Corey, Hurley, & Heilman, 2001). Additionally, abnormal patterns of cerebral gyri in the regions of frontal speech- and language relevant areas close to the lateral sulcus were found in further studies with adult PWS (Cykowski et al., 2008; Foundas et al., 2001). Altogether, it seems that in PWS the area surrounding the lateral sulcus, i.e. the perisylvian region, seems more heterogenous than in non-stuttering persons (Foundas et al., 2004). Furthermore, structural differences in the right caudate, leading to an atypical asymmetry (left warded), was found in CWS aged eight to thirteen years (Foundas, Cindass, Mock, & Corey, 2013). The caudate was measured with volumetric MRI scans in fourteen CWS and in a control group, displaying a reduction of volume in the right caudate and a subsequent asymmetry within the hemispheres compared to the control group. Authors concluded that those anomalies could cause a vulnerability for perturbations during speech planning and therefore result in an inefficient coupling of action and perception and a susceptibility for stuttering (Foundas et al., 2013).

Matter-reduction in the following left hemisphere areas may indicate that exactly those areas are relevant for the primary pathology of stuttering and are thus the most important morphological correlate: a reduction of gray matter correlating positively with stuttering severity in adult PWS and in recovered PWS was found in the left gyrus frontalis inferior. In CWS and children who recovered from stuttering, this reduction of gray matter was found to be located bilaterally in temporal areas (Chang, Erickson, Ambrose, Hasegawa-Johnson, & Ludlow, 2008; Chang, Zhu, Choo, & Angstadt, 2015; Kell et al., 2009). Replicable results were demonstrated with diffusion tensor imaging (DTI), determining the course of axons with fractional anisotropy. A reduction of functional anisotropy and, hence, a reduction of directionality, integrity and density in white fiber tracts of the left Rolandic operculum, located below the left hemispheric sensorimotor region for face, larynx and further organs relevant for articulation, was not only found in adult PWS but also in older children affected by stuttering (Chang et al., 2008; Connally, Ward, Howell, & Watkins, 2014; Cykowski, Fox, Ingham,

Ingham, & Robin, 2010; Sommer, Koch, Paulus, Weiller, & Büchel, 2002; K. E. Watkins, Smith, Davis, & Howell, 2008). Those fiber tracts are mostly part of the arcuate fasciculus (AF) and of the fasciculus longitudinalis superior (FLS), linking auditory cortical areas located in the superior temporal gyrus (STG) with frontal cortical areas. Therefore, they are part of the dorsal stream (Hickok & Poeppel, 2004, 2007). A further study by Cykowski and colleagues (2010), also using DTI in an adult PWS and fluently speaking persons, found results quite similar to Sommer and colleagues (2002): neuroanatomical aberrations were mostly restricted to the left hemisphere and found within the fiber tracts of white matter. In detail, it was also the FLS that was affected most by those aberrations. Cykowski et al. (2010) postulated the theory that one of the causes for stuttering might be the delayed myelinization of nerves within this stream, leading to an inefficient transport of information. This idea of the "delay in the myelination of the cortical areas in the brain concerned with speech" (Karlin, 1947, p. 319) emerged as early as 1947. According to this theory, an incomplete myelinization would therefore lead to a slower, less precise and more vulnerable transport of information and might thus also explain why emotionally or linguistically complex situations tend to increase disfluencies, since this path affected is close to areas of emotion or language, also located within the left frontal cortex. Interferences with activations in those areas might disturb information transport in process within the FLS as visualized in figure 2.3 (Guitar, 2014; Johnson, Walden, Conture, & Karrass, 2010).

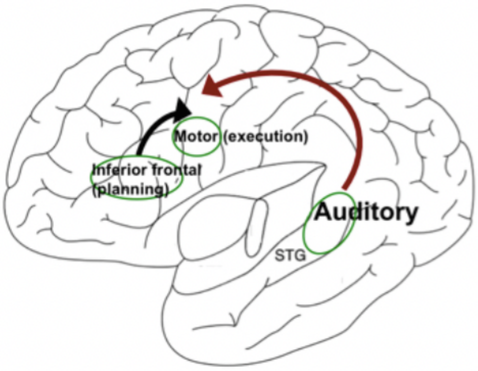


Figure 2.3 Simplified depiction of the left hemisphere
(featuring the inferior frontal regions for speech planning as well as the speech-motor cortex, which is relevant for the speech execution and the interconnection by the FLS, as depicted in red, Chang, 2011, p.5).

A common dysfunction in stuttering is located in the cortico-basal ganglia-thalamo-cortical loop, a complex loop from cortical areas leading to the basal ganglia (BG) and the thalamus and then back to the cortex of the frontal lobe (Giraud et al., 2008). Here, a reduction in functioning white matter in temporal areas as well as a reduction of gray matter in the left medial frontal gyrus and in temporal areas was demonstrated (Lu et al., 2010). Furthermore, regions with a reduced anisotropy were found in the corpus callosum of adult PWS: those anomalies, persisting through different processes of development, indicate a maladaptive reduction of interhemispherical inhibition, potentially associated with an unfavorable recruitment of right-hemisphere, frontal areas of the cortex, which are relevant for speech production (Civier, Kronfeld-Duenias, Amir, Ezrati-Vinacour, & Ben-Shachar, 2015; Civier, Tasko, & Guenther, 2010). However, a reduced fractional anisotropy was found in fibers linking auditory and motor regions as well as in fiber tracts linking cortical and subcortical areas and in the corpus callosum - not only in adult PWS but also in CWS (Chang et al., 2015). A further DTI finding that was replicated multiple times is the disrupted connection between the fiber tracts within the tractus corticospinalis, part of the pyramidal tract, and the effector motor neurons in CWS (Chang et al., 2008) as well as in adult PWS (Cai et al., 2014; Connally et al., 2014; K. E. Watkins et al., 2008). However, it has to be noted that in Watkins et al. (2008) salient results were surveyed and found in the right hemisphere, whereas in Connally et al. (2014), results were surveyed and found in the left hemisphere and in case of the latter, a further structure of the pyramidal tract, the tractus corticobulbaris, was functionally involved (Connally et al., 2014). A further recent study by Kronfeld-Duenias and colleagues (2016) found an elevated diffusivity which was negatively correlated with speech fluency in the frontal aslant tract in adult PWS. The frontal aslant tract is a neuronal path that was newly identified as belonging to the motor path of speech production, linking the inferior frontal gyrus with the supplementary motor area (SMA) and the premotor SMA. Those findings provide new and relevant information regarding the role of this tract in speech production and the etiology of stuttering.

Besides matter reduction, also an <u>increase in matter</u> was also found in various studies concerning specific neural correlates. An increase in grey matter within the basal ganglia, especially in the putamen, hints towards structural adaptions due to a proven dysfunction in those areas of PWS (Kell et al., 2009; Lu et al., 2010). Furthermore, an increase in white matter was identified in regions below the left hemispherical frontal regions as well as in right hemispherical temporal and frontal regions for speech planning, speech motor skills and auditory regions (Jäncke, Hänggi, & Steinmetz, 2004; Kell et al., 2009). Using voxel-based morphometry, Choo and colleagues (2011) demonstrated an increased white matter in the rostrum and the anterior midbody as well as an overall larger callosa area in adult PWS. This structure was also found to have aberrant fiber structure in children (Chang et al., 2015). Those results could point to anatomical changes in PWS that are associated with a different hemispheric distribution of processes relevant for language.

All studies cited above were conducted with adolescents or adults several years after stuttering onset, leading to a possible combination of causal and compensatory phenomena in the ontogenetic development. Altogether, four anomalies of the left hemisphere have proven to be central for developmental stuttering: First of all, it is the deficits in white fiber tracts below orofacial motor regions, secondly a reduction of grey matter in inferior frontal regions, including Broca's area. Both areas are part of the anterior cortical perisylvian areas relevant for speech and language and functionally connected during speech production. Therefore, exactly those anomalies in matter, highly important for the integration of articulatory planning and auditory feedback and for the execution of articulatory movement, might represent the primary lesions in stuttering. Increase in grey or white matter is a compensatory consequence in variable regions, especially the frontal, parietal and temporal ones (Neumann, 2007; Neumann & Euler, 2010). Since no augmentation in right hemisphere regions of speech and language and no aberrant symmetry was found in stuttering or recovered children (Chang et al., 2008), an increase in volume of right hemispherical structures in adult PWS might derive from persistent stuttering. The third anomaly of the left hemisphere that seems most relevant in neuroanatomical features of persistent developmental stuttering is in the corpus callosum as an interhemispherical link and the fourth is the integrity of fibers in the tractus corticospinalis.

However, it was not only differences in brain structure that were found in children, adolescents and adults who stutter but also in brain function. Those findings are explained in the following section.

2.5.2.2 Neurofunctional evidence

First neurophysiological results in stuttering research derive from electroencephalographic studies. It was already in those first studies that an abnormal laterality for usually strictly left hemispherical speech processes became evident (Moore, 1984a, 1984b; Moore & Haynes, 1980; Wells & Moore, 1990). Since the 1990s, functional imaging techniques have been essential for the understanding of functional consequences of stuttering on the brain and their link with the depicted morphological changes (see chapter 2.5.2.1). They detected neurofunctional correlates of stuttering in frontal and prefrontal regions of planning and execution of speech motor skills, as well as in regions of language and auditory processing and limbic and subcortical regions (Braun et al., 1997; S. Brown, Ingham, Ingham, Laird, & Fox, 2005; De Nil & Kroll, 2001; De Nil, Kroll, Kapur, & Houle, 2000; De Nil, Kroll, Lafaille, & Houle, 2003; Fox et al., 1996; Loucks, Kraft, Choo, Sharma, & Ambrose, 2011; Neumann et al., 2003; Neumann et al., 2005; Preibisch et al., 2003; K. E. Watkins et al., 2008). According to a meta-analysis by Brown and colleagues (2005), speech of PWS compared to PWNS is characterized by an extensive over-activation of motor regions, an anomalously right-shifted lateralization of activation in the frontal operculum, in the Rolandic operculum as well as in the anterior insula and by a missing auditory activation (in PWS only). Those aberrations and shifts of laterality are depicted in the following paragraphs.

In PWS, neuroimaging techniques have demonstrated functional and structural disruptions of connectivity within the basal ganglia (especially caudate nucleus, substantia nigra, putamen) as well as in the cortico-striato-thalamo-cortical loop multiple times. It is relevant for the control of executive functions like motor selection, sequential planning, affect, impulse and anticipation (Giraud et al., 2008; Kell et al., 2009; Lu et al., 2010). Additionally, new functional MRI meta-analyses showed that stuttering is associated with a reduced activation in left-hemisphere fronto-parieto-temporal regions (Belyk, Kraft, & Brown, 2015; Budde, Barron, & Fox, 2014). These findings are supported by results of a study on transcranial magnetic stimulation reflecting the cortical dynamics of local excitatory and inhibitory regulations and a reduced neuronal speech-planning dynamic concerning the primary motor cortex in stuttering speakers (Neef, Anwander, & Friederici, 2015).

A further non-invasive method, the high-resolution magnetoencephalography, also allows the detailed measurement of ongoing brain activity. It was used to investigate the cortical activation during speech production and showed a disturbed sequence in stuttering speakers: while in PWNS the left inferior frontal cortex, relevant for articulatory planning, was activated prior to the left motor cortex, which prepares the excitation of articulatory muscles, this sequence was reversed in PWS (Salmelin, Schnitzler, Schmitz, & Freund, 2000). A possible explanation for this phenomenon could be a functionally incorrect connection between left-hemispherical sensorimotor and frontal cortical areas, especially of Broca’s area, as it was structurally proven through DTI results named above (Chang et al., 2008; Kell et al., 2009; Sommer et al., 2002; K. E. Watkins et al., 2008). Hence, the neuronal communication between left-hemisphere regions of speech motor planning and execution, on the one hand, and auditory regions, on the other hand, seems impaired, maybe due to structural deficits in these regions (Neumann et al., 2005). This deficit is accompanied by a missing integration of auditory feedback of a speaker’s own speech into speech motor planning, which becomes clear when looking at the following facts: firstly, a disrupted inhibition of left-hemisphere auditory activity during speech, secondly functional and structural reorganization of right-hemisphere auditory areas through an increase of tonotopically (representation of a certain sound frequency in a certain area) organized right-hemisphere cortical areas, third, an increased volume of grey matter in the right gyrus temporalis superior (Beal et al., 2010; Kikuchi et al., 2011) and fourth, a presumedly reduced prevalence of stuttering in persons with a severe hearing disorder or deafness (Montgomery & Fitch, 1988). Another magnetoencephalography study demonstrated a changed hemispherical laterality with an abnormal right-hemispherical activation in the Rolandic operculum during speech perception in PWS (Biermann-Ruben, Samelin, & Schnitzler, 2005).

A disrupted temporal processing and an abnormal lateralization in right-hemisphere networks also in non-speech motor tasks, like finger-tapping (Max & Yudmann, 2003; Neef, Jung, et al., 2011) or during transcranial magnetic stimulation of orofacial motor areas, that are followed by an aberrant, right-hemisphere pattern of inhibition (Neef, Paulus, Neef, von Gudenberg, & Sommer, 2011) hint strongly towards the fact that stuttering may not only affect speech motor skills but also other motor functions when including right-hemisphere areas. This knowledge is important with respect to the studies conducted in the thesis at hand.

When investigating the effect of stuttering therapy (like fluency shaping) or recovery from stuttering on a neurological level, it was evident that neuronal processes of reorganization had taken place: an increased activity in frontal motor areas of speech and language as well as in temporal regions was found. Moreover, a certain shift of activation in the left hemispherical regions, especially those regions close to anomalies in the fibers of the AF and adjacent fiber tracts, was demonstrated (De Nil & Kroll, 2001; Neumann et al., 2003; Neumann et al., 2005). This was further confirmed and specified by recent functional MRI meta-analyses: an increase in speech fluency was accompanied by an enhanced co-activation of right hemisphere fronto-parieto-temporal areas (Belyk et al., 2015; Budde et al., 2014). Furthermore, a post-therapeutic normalization of function within the BG, the anterior insula and the auditory cortex bilaterally was observed (Giraud et al., 2008; Neumann et al., 2003). Each of these regions is involved in the integration of sensory and auditory feedback into the motor system, which seems to be of prime importance for stuttering therapy and its success (Kell et al., 2009). For the cerebellum, too, a normalization of a pre-therapeutic increased functional connectivity during rest was achieved by a reduction of this increased connectivity to the level of PWNS after a successful and effective stuttering therapy (Lu et al., 2012). Principles of neuronal plasticity after brain lesions are for example the expansion of active areas and the usage of homologous areas in the other hemisphere (Grafman, 2000). The observed over-activation in treated and untreated PWS (S. Brown et al., 2005) shows a compensatory expansion and an involvement of Brodman area 47 (in the right hemisphere), which is claimed to be the homologous area of Broca’s region. This is stated to be an adaption of homologous areas to the function of contralateral regions. An effective therapy, however, translocates the compensation for deficits within the left hemisphere from right-hemisphere, homologous areas into left-hemisphere, perilesional regions. Therefore, an effective compensation seems to demand the restauration of left hemispherical networks, similar to aphasias caused by strokes (Heiss, Kessler, Thiel, Ghaemi, & Karbe, 1999; Rosen et al., 2000). This way, speech fluency inducing conditions, using a slow and uniform speech rhythm, can operate as an external pacemaker and thereby synchronize the disturbed flow between auditory areas and areas of speech motor planning and speech motor execution (Neumann et al., 2005).

In PWS who did not undergo therapy, regions and extent of brain activity correlating positively with stuttering severity could be attributed to a functional cause for stuttering, whereas regions with a negative correlation might have a rather compensatory function. A negative correlation of over-activation in the right orbitofrontal cortex (Brodman area 47) with stuttering severity in untreated PWS (persons stuttering less severely activated this region more than persons stuttering heavily) implies a compensatory function of this region (Neumann et al., 2005; Preibisch et al., 2003). A compensatory role of brain regions within the intact hemisphere is also described in strokes. In PWS, however, this spontaneous compensation seems to be insufficient, since the person affected continues to stutter (Heiss et al., 1999).

In a study by Kell et al. (2009), a positive correlation for brain activity and stuttering severity in left perisylvian regions (anterior insula, Rolandic operculum), bilaterally in the auditory cortex (planum polare) and the striatum (part of the BG) was found, hinting towards a causal pathology in those areas. As the correlation between stuttering severity and brain activity completely disappears after a successful therapeutic intervention leading to speech fluency, this area is a suitable goal area for a therapy. This effect was demonstrated in the left anterior insula as well as bilaterally in the planum polare and the striatum (Kell et al., 2009). In contrast, a positive correlation persisted between stuttering severity and brain activity within the primary motor areas of articulation in the left Rolandic operculum. The same is true for a negative correlation in the right Brodman area 47/12 (orbitofrontal cortex), indicating a post therapeutic persisting pathological function of the first region and an ongoing compensatory function of the latter region. This functional MRI investigation comparing recovered stutterers with PWS and with PWNS detected that the only over-activated region was an area in the left orbitofrontal cortex located exactly within the homologous area of the right region of compensation (Brodman area 47/12). This area comprises linguistic, motor and (rhythmical) speech motor functions and seems to eliminate disfluencies through the adaption of discrepant speech metrical structures into an adapted motor program. Hence, this exact region could be the primary goal structure for a long-term success of therapeutic intervention. Altogether, effective therapeutic interventions should aim towards a re-functionalization of orbitofrontal, auditory and basal ganglia regions of the brain (Kell et al., 2009).

All studies cited above were conducted with adolescent or adult PWS. One study investigating very young CWS is mentioned, since data on young children still in the phase of developing stuttering bring extremely relevant information regarding causal aberration on neurological levels. Sowman and colleagues (2014) investigated hemispheric dominance in pre-school CWS and in fluently speaking peers using magnetoencephalography. Children were asked to participate in a picture naming task, while their brain activity was being recorded. Results showed that activation patterns in speech relevant areas did not differ between groups. Activation was located within the left hemisphere and therefore shows the opposite of studies investigating adult PWS (Sowman, Crain, Harrison, & Johnson, 2014). The authors therefore concluded that a changed hemispheric dominance is more likely to be of compensatory nature (i.e. neuroplastic adaption) than of causal nature during the process of manifesting stuttering.

All in all, those studies investigating the neurofunctional evidence of developmental stuttering showed various aberrations in form of over- or under-activation within certain areas of the brain in PWS. Namely, there was an over-activation found in PWS in fronto-parietal motor areas specifically within the right hemisphere, as well as in left hemisphere areas of the cerebellum. An over-activation was also found during speech in the right orbitofrontal cortex. Simultaneously, a lower activation was found within the left fronto-parieto-temporal areas during speech as well as in auditory areas during stuttering, which was reversed into an over-activation during fluent speech. Those results raise the question as to how auditory monitoring influences speech fluency. Some researchers claim that monitoring enables a synchronization or integration of a specific sequence of activities. This process is running parallel to the decision of a speaker about what he/she wants to say and how to articulate it. An asynchrony or a timing deficit could therefore be considered one of the causes for stuttering (Guitar, 2014; Van Riper, 1971). Additionally, it was found that the left motor cortex is activated prior to the left inferior cortex. This indicates a reversed order because the first is relevant for the triggering of articulatory muscles, whereas the latter is relevant for articulatory planning. However, those patterns can also be changed since the condition after successful therapy or after recovery from stuttering as well as the investigation in very young CWS show different patterns i.e. an increased and expanded activation in areas associated with speech and language compared to the pre-therapeutic status, a normalization of speech-associated activation in right and left hemisphere areas and a re-functionalization of the BG during speech. Furthermore, an increased activation in auditory areas was demonstrated during fluent speech. An activation within the left orbitofrontal cortex and in the auditory cortex bilaterally was found after a complete recovery from stuttering. Those results after effective therapy or recovery all point towards a normalization of neurofunctional activation, comparable to fluently speaking persons.

2.5.2.3 Summary of recent neurological findings

The primary goals of the last two sections were to give a comprehensive overview over recent findings in research and to focus on results that provide relevant information for the thesis at hand. It does not suffice to look at aberrations in anatomy and in function, the causal and compensatory factors in developmental stuttering also need to be considered. Whilst aberrations and anomalies can be captured by neuroimaging techniques, the question whether the observed aberrations are to be seen as a cause or a consequence of stuttering is still not completely answered, although tendencies are becoming clearer. Comparing all studies cited above, one must be cautious when interpreting their findings, since neither methods nor participants can be compared directly. However, all those results can be reviewed and give us an idea concerning the question of cause versus compensation.

Stuttering persons differ from fluently speaking persons regarding their neuroanatomy: a reduced density and an aberrant directionality in specific fiber tracts of the left hemisphere Rolandic operculum lead to an interruption of the integration of sensorimotor information. Those changes observed in the fiber structure might have been caused by an incomplete maturation process. Furthermore, PWS show less volume of grey matter in specific areas, namely within Broca's area and the adjacent regions. Results on neuroanatomical differences could also be replicated in children.

Those structural changes are followed by functional aberrations and a connection between them can mostly be made directly. In the case of neurofunctional findings, over-activations or under-activations in certain areas were demonstrated. Areas of the right hemisphere frequently showed over-activations in cortical as well as in subcortical areas, with the main difference regarding the activation in the Rolandic operculum and the anterior insula as well as the basal ganglia. A minor activation was found in all areas of the ventral PM, the opercular cortex and sensorimotor areas of the cortex in both hemispheres as well as in the auditory areas bilaterally, especially within the temporal regions. In pre-school children however, those findings of a changed hemispherical dominance could not be replicated, leading researchers to conclude that a changed laterality is a consequence rather than a cause of stuttering.

Hence, numerous anatomical differences can be found that are associated with an aberrant function. Developmental stuttering is therefore accompanied by genetically caused morphological and functional aberrations compared to fluently speaking persons, with networks of speech, language and auditory processing being particularly affected. Those differences are apparent in nearly all age groups. Solely young children do not seem to display a changed neurophysiology. Furthermore, persons who recovered from stuttering or lost their disfluencies due to an effective therapeutic intervention also show comparably normalized patterns of neurofunctional activation. Even though current data has brought much new information to the neuroanatomical and neurofunctional basis of stuttering, further research is needed to get closer to the connection of cause and consequence in stuttering. After this comprehensive summary, models of speech processing in stuttering and their relevance in explaining the phenomenon are discussed.

2.5.3 Models of speech processing in stuttering

Neuroscience hypotheses of the emergence of stuttering encompass a changed structure of hemispherical dominance, a dysfunction of the basal ganglia, a disconnection syndrome, changed cerebral timing-networks and a disrupted sensorimotor integration. All of those aspects seem reasonable, given the complex and interwined cognitive, linguistic and sensorimotor processes relevant for speech production (Guitar, 2014; Neef et al., 2015). The generation of fluent speech is dependent on ongoing dynamic interactions between auditory, somatosensory and speech motor networks. Considering those sensitive interactions, it is hardly surprising that even minimal interruptions within those networks can have massive impact on speech motor output. Therefore, research has not only tried to capture neurological groundings of stuttering but it has also attempted to create models of stuttering and its loci of dysfunction. Those models shall be explained in the following. However, since a comprehensive explanation of all popular models is not possible in the thesis at hand, it focuses on the most important and – for the research questions to follow – most relevant ones.

Modern models of stuttering assume a disturbed motor control (Civier, Bullock, Max, & Guenther, 2013; Civier et al., 2010; Max, Guenther, Gracco, Gosh, & Wallace, 2004) and consider it a problem in the execution of highly complex and automatized sequences of motor commands as well as a problem of vulnerability within the senso-motor system, which seems reasonable with regard to the extremely high tempo needed in fluent speech planning and production. It was as early as in 1950 that Holst and Mittelstaedt postulated the so-called *reafference principle,* a model or feedback control system not specific to stuttering but rather generalized for all self-initiated motions (Holst & Mittelstaedt, 1950). It controls motion sequences by sending a movement instruction (efference) to the effector organs (i.e. articulatory muscles). A kind of copy (efference copy, a neural representation of motor outputs) is saved temporarily within the central nervous system as a representation of the actual goal. Effector-organs and sensory organs send feedback (re-afferences) about the success of the movement, which is then compared to the efference copy. In case of deviations from the copy, higher levels of the central nervous system keep on correcting until feedback and efference copy are as similar as possible. According to an integrative scheme of motor control, movements are being controlled by feedforward and feedback systems. The feedforward projection of the motor plan, the efference copy, is sent to the sensory system, where the perception of the planned movement is expected. Consequently, efference copies are used by the feedback system to compare the expected result with the actual movement. This principle is still valid today and integrated into various newer models.

Researchers, especially in the circles around Max and colleagues (2004) and Civier and colleagues (2010, 2013), presume that PWS speak disfluently due to a shift of feedforward to feedback control during speech motor processes combined with an exaggerated dependency on a slow and disturbed auditory feedback control system. These aberrant processes are likely to lead to mistakes in speech production, that – if big enough – end in a reset of the speech motor system by repeating syllables (Civier et al., 2010). This assumption is part of the DIVA (Directions into Velocities of Articulators, see figure 2.4) model of speech production, that was later expanded to the GODIVA (Gradient Order DIVA) model. The latter explains stuttering and its removal by simulations of these processes and also the fluency inducing-impact of slowed and prolonged speech as well as masking or shadowed speech (Civier et al., 2013; Civier et al., 2010; Guenther, 2016; Tourville & Guenther, 2011). The DIVA model is one of the most comprehensive neurocomputational models of speech motor control and a “hybrid control system combining a model-predictive controller with separate auditory and somatosensory feedback controller loops” (Parrell, Lammert, Ciccarelli, & Quatieri, 2019, p. 1463). To be more precise, the model depicts an adaptive network for the three components of speech, namely acoustic, somatosensory and, of course, motor components of speech. Numerous cortical and subcortical areas involved in speech are considered as well as the connections between those areas. The model uses its feedforward and feedback systems to reach four different goals: to engineer speech production, to collect input about acoustic features of speech and the sensory features of involved oro-facial, vocal tract or other related muscles, to make comparisons between the sounds produced and to adjust according to learned templates. The DIVA model starts with the speech sound map and explains processes of phonetic encoding and articulation. It is also the speech sound map as well as the articulator map controlling the articulatory muscles where the feedforward and feedback mechanisms converge. Frequently spoken phonemes, syllables and words are included in the speech sound map, which also encodes the following motor, auditory and somatosensory programs of each specific speech sound. These programs define which sensory signal to expect and consist of sets of neurons with their axonal projections generating a known sequence of articulator movements. Subcortical loops control projections from the speech sound map: in the feedforward control system, it is the cortico-basal ganglia loop that controls the initiation of the following speech sound and inhibits the preceding speech sound. Within this loop, the sensorimotor and cognitive components work together to ensure the correct sequencing of speech sounds. After the activation of a speech sound via the cortico-basal ganglia loop, motor and sensory programs start the encoding process. The auditory and the somatosensory subsystem detect and correct aberrations between their programs and the actual sensory state. Whenever a deviation occurs, sensory error maps immediately activate the feedback control map to correct the motor command in the articulator map. The feedback control system encompasses those two subsystems.

The expanded version, the GODIVA model was extended by a planning and a motor loop. Simulations of disfluencies demonstrated a significant role of the BG, the thalamus and the left ventral premotor cortex (PMC). Two hypotheses have been tested, one claiming that white-matter abnormalities lead to a disturbance in the corticostriatal circuit, the other that dopaminergic aberrations lead to disturbances in the circuit passing the striatum. Results of the simulation suggest that both scenarios are plausible, since a delayed readout of the following syllable's motor program was observable in both cases, leading to a disfluency as observed in stuttering. Furthermore, those results confirm findings of brain imaging during stuttered speech. Therefore, authors come to the conclusion that both abnormality types can lead to stuttering moments that are most likely caused by interferences of the BG-thalamus-ventral PMC circuit (Civier et al., 2013). Abnormalities within the auditory feedback loop explain why a changed auditory feedback of one's own speech in PWS can lead to speech fluency (whilst in PWNS the exact opposite happens). Movements of the same effect organ (i.e. sucking or chewing) are not affected (Civier et al., 2013; Civier et al., 2010; Guenther, 2016; Max et al., 2004; Tourville & Guenther, 2011).

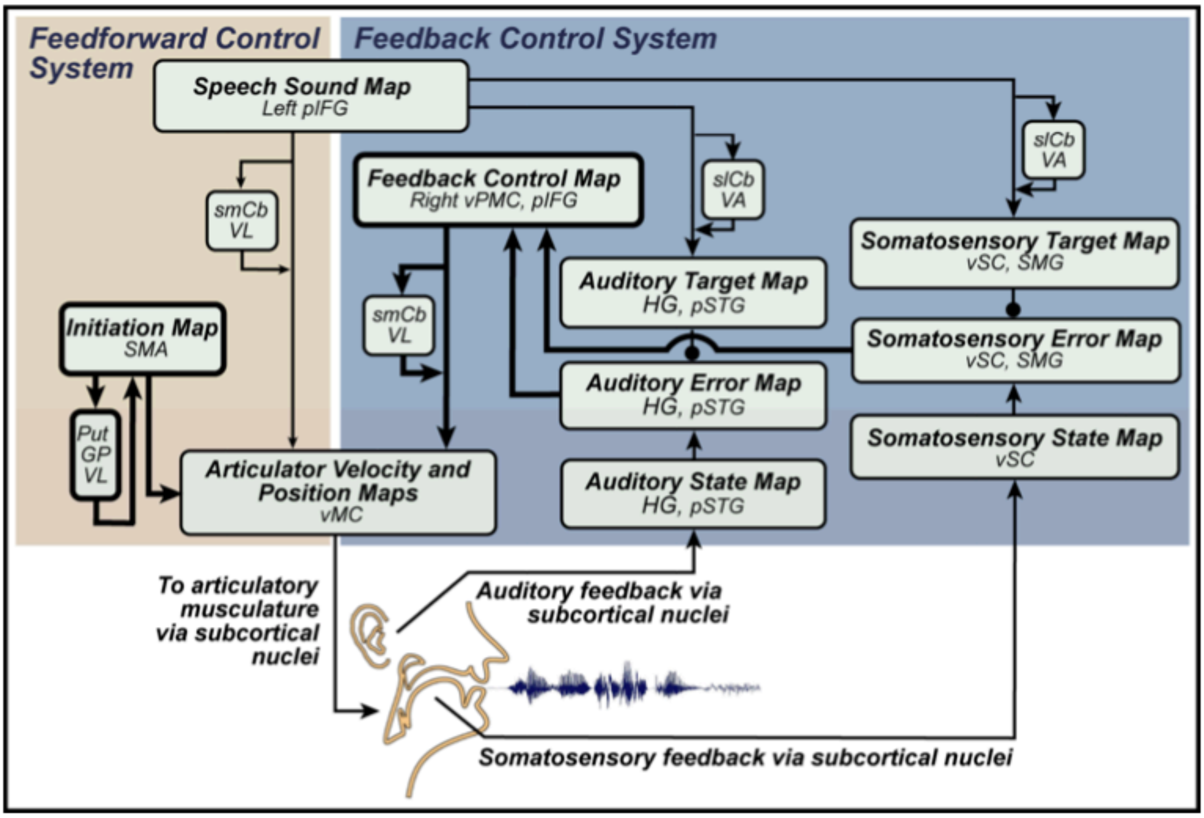


Figure 2.4 The DIVA model of speech acquisition and production by Tourville and Guenther (2011, p.23).
GP, globus pallidus; HG, Heschl's gyrus, pIFG, posterior inferior frontal gyrus; pSTG, posterior superior temporal gyrus; Put, putamen; sLCb, superior lateral cerebellum; smCb, superior medial cerebellum; SMA, supplementary motor area; SMG supramarginal gyrus, VA, ventral anterior nucleus of the cerebellum; VL, ventral lateral nucleus of the thalamus; vMC, ventral motor cortex; vPMC, ventral premotor cortex; vSC, ventral somatosensory cortex;

Speech processing is based on extensive, dynamic networks in which cortical, subcortical, and cerebellar areas interact. To understand those systems, it is important to know the regions involved and the anatomical connectivity between them. Integrity and density of cerebral fiber tracts, enabling a functional interaction, are essential to this (Dick, Bernal, & Tremblay, 2014). Functional connectivity based on functional MRI investigations describes connections between two systems that can either be linked directly or indirectly via a third system. Structural connectivity, investigated by DTI and various procedures of visualization (i.e. tractography, sectional images etc.), document the morphological connections within those systems. The knowledge of a functional connectivity allows the implementation of a causal model of functional connectivity, visualizing the influence of one neural system to another (Guenther, 2016).

A modern model of auditory speech processing assumes two interacting streams or paths of cerebral architecture, one dorsal and one ventral one, that are grounded in long fiber tracts, to which the AF/FLS belongs: it is called the *dual-stream model* (Hickok & Poeppel, 2004, 2007):

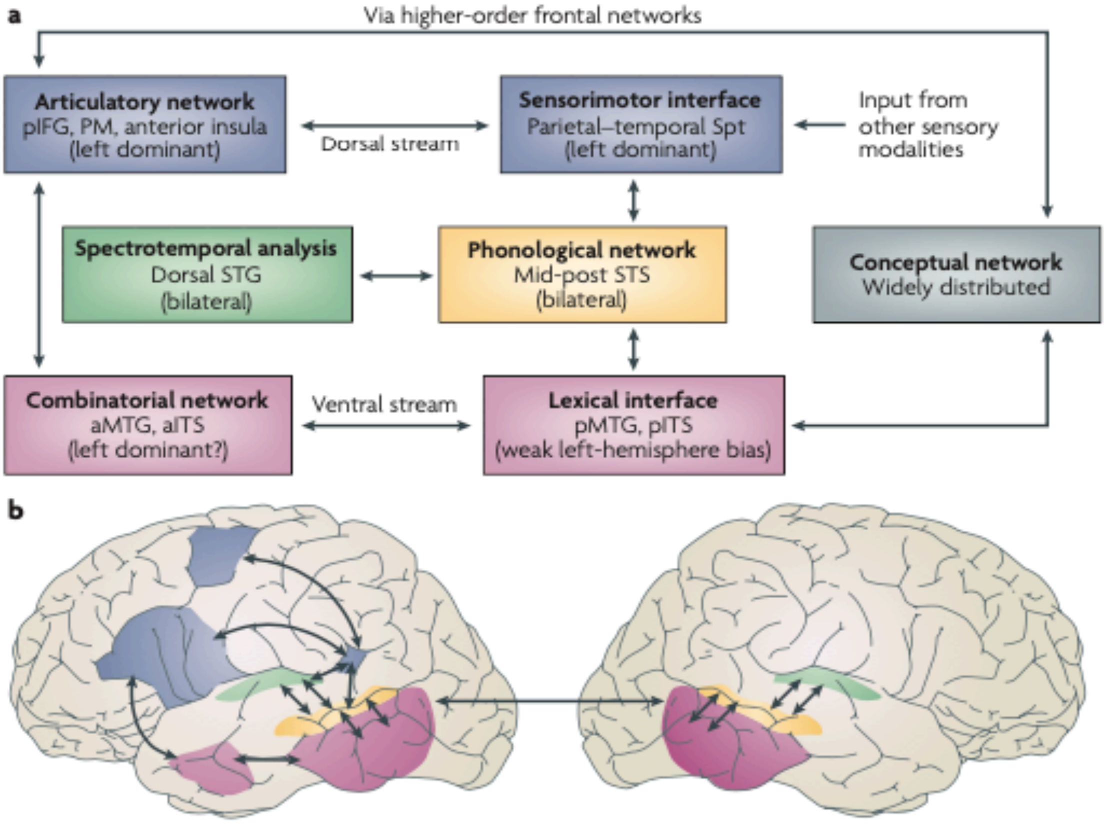


Figure 2.5 Graphic of the dual stream model containing a) a schematic diagram and b) a estimated anatomical locations of the model (Hickok & Poeppel, 2007, p. 395).
pIFG, posterior inferior frontal gyrus; PM, premotor cortex; STG, superior temporal gyrus; STS, superior temporal sulcus; aMTG, anterior middle temporal gyrus; aITS, anterior inferior temporal sulcus; pMTG, posterior middle temporal gyrus; pITS, posterior inferior temporal sulcus;

At first, there are joint steps of cortical speech processing involving a kind of spectrotemporal analysis in the STG (auditory cortices), which is involved in those early stages of cortical speech perception. Due to differences in the spectrotemporal computations and in demands between the hemispheres, the system splits into two streams: one is the dorsal pathway depicted in blue and mapping phonological or sensory representations to the articulatory representations, the other is the stream depicted in purple, the ventral pathway, mapping phonological or sensory representations to the lexical representations (Hickok & Poeppel, 2004, 2007). The dorsal path is, therefore, relevant for the depiction of speech sounds within the articulatory networks of the frontal lobes, meaning that the sensorimotor integration of the perceived own speech into motor speech planning is happening here. According to the authors, for the comprehension of speech, a speech sound has to be linked to an articulatory representation. Since there is no one-to-one-correspondence of a spoken sound to the perceived signal, speech sounds have to be represented in their invariant, i.e. their motor form, within the brain. It is therefore necessary for a precise articulation to get feedback from the auditory-motor integration loop. The ventral path is projecting bilaterally ventrolaterally to temporal cortical areas and is thus used as an interface of sound-to-meaning by mapping speech sounds to conceptual representations (assignment of meaning to a simple acoustic signal). When looking at the anatomical locations of the dual-stream model in detail, regions shown in green stand for areas on the dorsal parts of the STG. Areas in yellow located in the posterior half of the superior temporal sulcus are relevant for phonological processes. Regions kept in purple are part of the ventral stream, that has a slight left-hemisphere bias with a generally bilateral organization. Areas in blue depict the dorsal stream, which is located within the left hemisphere. There is a correspondence of posterior regions of the dorsal stream with a location within the sylvian fissure, more precisely at the parieto-temporal boundary, that is claimed to be a sensorimotor interface. The rather anterior locations within the frontal lobe, that probably involve Broca's region are proposed to be portions of the network relevant for articulation (Hickok & Poeppel, 2007). Stuttering is mostly considered a problem of speech motor control and less a linguistic problem. It therefore should mostly affect dorsal paths, which has been demonstrated multiple times (Kell et al., 2009; Kronfeld-Duenais, 2014). By using tractography, Kronfeld-Duenias and colleagues (2016) demonstrated reduced volumes in dorsal paths, namely within the AF and the anterior segment of the FLS. A meta-analysis concerning DTI results in stuttering (Neef et al., 2015) attested a reduced fractional anisotropy and therefore deficient fiber tracts in the dorsal stream and the interhemispherical connections between sensorimotor cortices. Furthermore, it seems plausible that a disrupted processing of internal feedforward models in stuttering within the dorsal stream leading to an impaired integration of one's own speech into the speech-motor planning and execution are in line with the fact that PWS have less reliable phonological percepts than PWNS, for example a reduced precision in the perception of certain stop consonants (Neef et al., 2012). Additionally, a successful stuttering therapy was shown to reduce the over-activation in dorsal regions, that are to be interpreted as compensation (Kell et al., 2009).

In sum, the generation of speech fluency demands ongoing dynamic interaction between auditory, somatosensory and speech motor networks. According to the dual-stream model, mostly dorsal paths relevant for the auditory-motor integration are affected in stuttering (which is considered a problem of speech motor control). This indicates a disrupted processing of internal feedforward mechanisms (i.e. projections of the motor plan that are sent to the sensory system to generate a perception according to the planned movement) and auditory feedback mechanisms (GODIVA-model) along with an impaired integration of one’s own perceived speech in the speech motor planning and execution as well as a disrupted BG-thalamus-ventral PMC circuit. Successful stuttering therapy in adult PWS has shown to reduce the over-activation of dorsal regions and lead to rather normalized activation patterns. Since both models have some factors in common and some factors are interpreted differently, one cannot be called superior to the other. Brain imaging and computational studies provide evidence for the plausibility of both theoretical constructs.

2.6 Stuttering and (speech) rhythm

The following chapter provides information about the role of rhythm for the perception, processing and production of speech and language. Initially, the question as to what rhythm exactly is and which role it might play for PWS shall be clarified. This theoretical basis is relevant for the research questions of the thesis at hand as well as the derivation of the hypotheses for the three studies to follow.

2.6.1 The relevance of rhythm for speech perception and production

The word *rhythm* descends from the Latin word *rhythmus* and the Greek word *rhythmos* and means *flowing* (A. C. Lewis, 2007). A general definition of rhythm contains the order of a temporal progress, a more speech-specific definition defines rhythm as the structure or organization of speech production that emerges from the alternation of long and short as well as stressed and unstressed syllables (Fujii & Wan, 2014; A. C. Lewis, 2007). Consistent forms of rhythm are classified as periodic rhythm since they appear as a reoccurring pattern. Both in speech and in music, temporal regularities exist at various hierarchical levels. Whereas in the case of music, temporal intervals of the perceivable beat are mostly isochronous and, therefore, lead to a more salient regularity, in speech this regularity is sometimes harder to perceive due to a higher variability (i.e. quasi-periodic) of intervals (Goswami, 2019; Ladány, Persici, Fiveash, Tillmann, & Gordon, 2020). The sense of rhythm is the ability to perceive this very temporal order. Every human is born with a certain sense of rhythm, everyone’s first experience with it being the heartbeat within the maternal body. The ability to perceive or to produce rhythm can still vary significantly between individuals (Fujii & Wan, 2014). It is part of human communication and social interaction. In spoken language, speech rhythm is embedded in the amplitude envelope: it contains information about duration, stress and the general tempo of speech (Ladány et al., 2020). The normal speech rhythm in most languages of the world is quite alike and amounts to three to eight syllables per second, which corresponds to three to eight hertz. This natural rate of velocity in speech is composed of a temporal sequence in interacting movements of articulators such as tongue, velum, jaw, lips, and vocal folds. A disruption or change of this fine-tuned rhythm can lead to misunderstandings, since the comprehension of speech is strongly dependent "on the integrity of its temporal envelope" (Ahissar et al., 2001, p. 13367). Information that is contained in this envelope is crucial for the identification of sentences, words, syllables and phonemes. In normal speech, frequency is mostly below eight hertz, ranging from four to 16 hertz. However, unimpaired brain mechanisms can adapt to varying input rates if they are within this range. This process of adaption is essential for speech perception, since speech rates can vary strongly between different persons and are also sensitive to changes caused by emotional states of the speaker (Ahissar et al., 2001; Fujii & Wan, 2014). Ongoing interruptions of the speech flow can lead to a decreased intelligibility, as it can be the case in stuttering. Rhythm, therefore, is essential for the perception and production of spoken language. The anatomical structures relevant for these processes will be explained in the following.

2.6.2 Neural correlates of rhythm perception and rhythm production in speech

Various studies have been conducted – for example using MRI technologies – to find out about the specific structures involved in the perception and production of speech rhythm. Due to the vast extent of the field and more detailed information in the three studies to follow, only the basic information is going to be presented in this chapter. A summary on the recent status of research shall be given based on a model of rhythm processing and rhythm production.

Concerning the perception of rhythm, the current state of research proposes that the temporal cortex gets its auditory input from the brainstem via the thalamus. The temporal cortex sends information to the prefrontal cortex, which is depicted with pink arrows in figure 2.6. It is presumed that these processes are located mainly within the right hemisphere. Also, the cerebellum receives input from the brainstem and passes this information on to the SMA. Here, again, the forwarding of information works via thalamus. The SMA as well as the prefrontal cortex transport this information to the BG, which subsequently forward the auditory signals via the BG-thalamo-cortical loop (marked in light blue in figure 2.6, part A).

To produce speech rhythm, the authors assessed that the primary motor cortex receives its input from the SMA which is part of the SMA-BG-thalamo loop (depicted with green arrows, part B of figure 2.6). Additionally, the primary motor cortex gets further input from the left hemisphere

PMC as well as the inferior frontal gyrus, transforming speech sounds into motor commands (as depicted with red arrows in part C, figure 2.6). The left PMC and the inferior frontal gyrus transfer the signals to the temporal cortex, which is responsible for the sensory prediction. The temporal cortex surveils the sensory predictions and compares them with the auditory feedback that is received from the brainstem, again via thalamus. Feedback errors of the temporal cortex are directed to the right hemisphere PMC and the inferior frontal gyrus, with both being interconnected to the thalamus as well as the cerebellum (as depicted with light blue arrows in figure 2.6, part B) (Fujii & Wan, 2014).

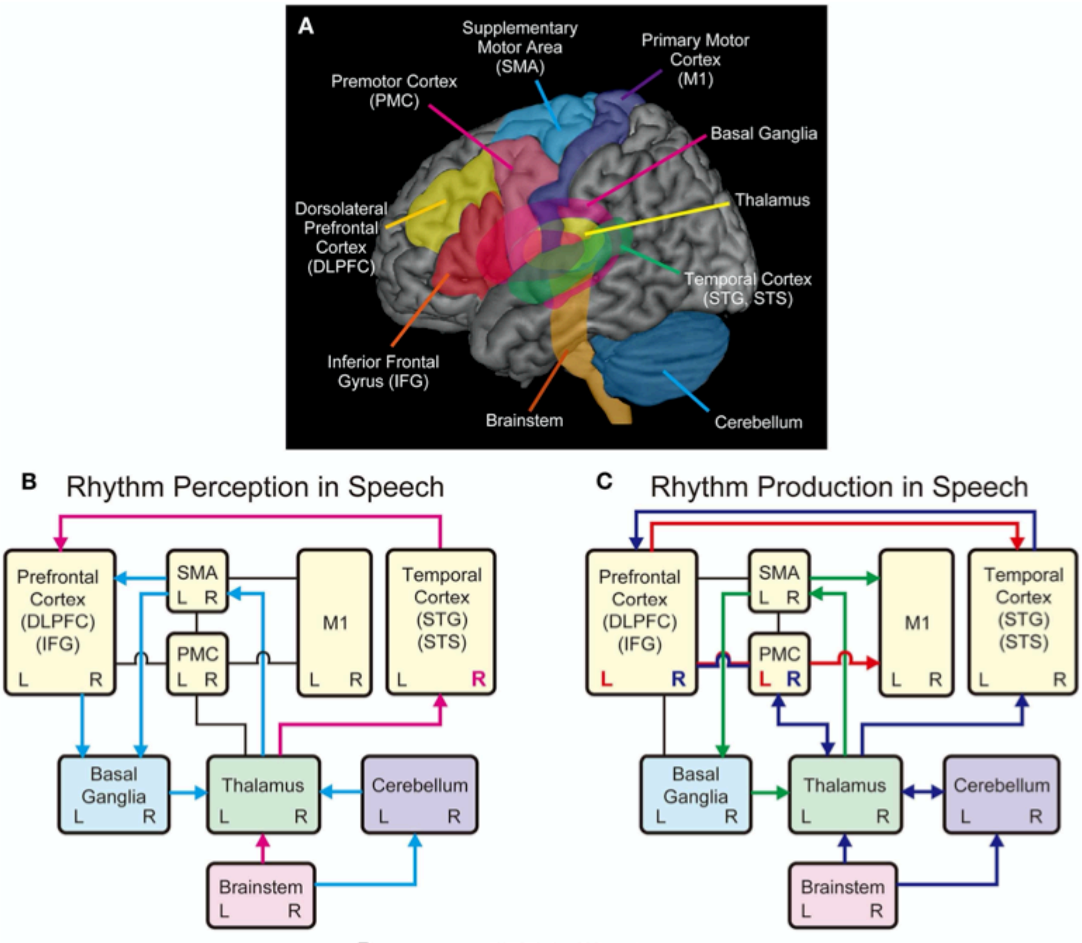


Figure 2.6 Schematic model of the network for the perception and production of speech rhythm (Fujii & Wan, 2014, p. 4).
DLPFC, dorso-lateral prefrontal cortex; IFG, inferior frontal gyrus; STG, superior temporal gryrus; STS, superior temporal sulcus; M1, primary motor cortex;

Comparing those structures for the perception and the production of speech rhythm with those that showed aberrations in PWS, it becomes evident that there is an overlap in some regions. This indicates that those exact structures relevant for the perception and production of rhythm show demonstrable aberrations in PWS. Structures showing this overlap are, first and foremost, the PMC in both hemispheres as well as temporal areas relevant for the processing of auditory stimuli and the BG (Cai et al., 2014; Fujii & Wan, 2014; Kell et al., 2009; Neef et al., 2015; K. E. Watkins et al., 2008).

2.6.3 Stuttering – a deficit in rhythm or timing?

The assumption that stuttering could be associated with a deficit in rhythm or timing has been discussed previously in the relevant literature. Different investigations from many years ago were able to demonstrate that PWS show more problems when it comes to the temporal coordination of movements than fluently speaking persons (M. Adams & Hayden, 1976; Borden, 1983). Since research questions today are going back to those results, a short depiction of those studies shall be given. Adams and Hayden investigated a hypothesis in 1976, proceeding from the assumption that PWS display more problems in the initiation and termination of phonation than fluently speaking control participants. To investigate this hypothesis, they tested ten adolescents who stuttered and ten fluently speaking adolescents. The task was to start or to stop phonation as quickly as possible when hearing a specific signal. Vocalization was recorded permanently and results showed that the group of stuttering participants was significantly slower regarding the initiation and termination in reaction to the signal. They were significantly inferior on most measures compared to the fluently speaking control group (M. Adams & Hayden, 1976). An investigation by Borden (1983) produced similar results. This study compared intervals in the initiation and the execution of fluent speech in PWS and fluently speaking persons during counting. Counting was done both with phonation, and silently (i.e. with only manual movement). Results demonstrate clearly that PWS with severe stuttering were significantly slower in silent and in aloud reading compared to the control group. Participants who stuttered mildly were comparable to control participants in their performance. In both tasks it was the execution of oral and manual movements that was responsible for this delay, the time for initiation only played a minor role (Borden, 1983).

Both studies therefore suggest that PWS display a motor timing deficit that also interferes with movements of speech and articulation, since it seemed to be the component of execution that was affected rather than the planning (Borden, 1983). Wing and Kristofferson (1973a, 1973b) also conducted a study on motor control abilities (finger tapping) in a fluently speaking population. Based on their results they postulated a model explaining motor timing deficits. The authors supposed that two distinct components are involved the ongoing processes: while the peripheral motor component is approximately the same in all individuals and therefore only accounts for a relatively small part of the overall variance, the so-called *central timekeeper* is assumed to be the main source of those individual differences concerning the variability and resulting timing deficits. According to this model it would not be the execution, as Borden (1983) suspected, but rather the central temporal planning that leads to deficits (Howell, 2004; Wing & Kristofferson, 1973a, 1973b). Another theory tries to further specify the timing deficit: the central clock hypotheses is based on the fact that PWS show a higher temporal variability in their speech production compared to PWNS. This hypothesis has been discussed by various authors so far (Cooper & Allen, 1977; Hulstijn et al., 1992; Zelaznik, Smith, & Franz, 1994). This increased variability becomes evident in patterns like the subglottal air pressure, utterance length, vocal duration and the voice onset time[2] (VOT). All of those parameters could be a manifestation of an instable central timekeeper (Guitar, 2014). It was furthermore observed that the probability of finding timing variability in PWS was higher in more complex tasks (Falk, Maslow, Thum, & Hoole, 2016; Falk et al., 2015; Howell, Au-Yeung, & Rustin, 1997; Hulstijn et al., 1992; Olander et al., 2010). Results of behavioral studies have demonstrated that stuttering persons were slower, less precise or temporally more variable in verbal and non-verbal tasks. At the same time, however, results in this domain are also inconclusive and heterogenous (Max & Yudmann, 2003).

Besides many studies on the motor component of the timing deficit in stuttering, there were other approaches, too. A theoretical model on the emergence of stuttering due to deficient auditory feedback processes was postulated by Harrington (1988) many years ago. The model of fluent speech production, from which the model of speech production in stuttering is derived, supposes that fluent speech production is mainly based on a system of rhythmical processes of integration. It contains a plan specifying relative timing. A good example is the case of a syllable that is composed of a consonant and a vowel: the speech production system must anticipate the interval in which the articulatory strategy for the production of a consonant transits into the articulatory strategy for producing a vowel. On top of that, the speech production system contains a rhythmical structure enabling the speaker to foresee the occurrence of vowels in a stressed syllable. This way, the speaker can structure his or her speech, since stressed syllables tend to reoccur in a predictable rhythm and can therefore be expected by the speaker. However, a successful prediction can only be made if the expectance of the speaker is compatible with the perception of speech signals. A deficit in the auditory perception of those signals can confound the whole system, since predictions suddenly do not seem to apply anymore. Based on this model of speech production in fluent speakers,

---

[2] The phenomenon happens in plosives and is defined as the time between the release of a stop consonant and the beginning of vocal fold vibration (Gick, Wilson, & Derrick, 2013).

Harrington postulated his theory of speech production in PWS: According to his theory, the deficit lies in the prediction of vowels in stressed syllables, since their auditory feedback processes rely on a dysfunctional perception that can be described as an anticipation. To illustrate this hypothesis, an analogy can be used: a passenger is waiting for an apparently late train, because the schedule implies that the train should have arrived already. Consequently, the passenger assumes a delay of the train. However, the schedule was wrong, and the train arrives on time – even if the passenger assumed the opposite. In this case an incorrect interpretation of events took place. According to Harrington (1988), this is exactly what happens in the speech production of PWS. Let us assume that the person plans to utter a syllable consisting of a consonant and a vowel and starts off with the consonant. The person then expects to perceive the production of the following vowel too early, since the actual production takes more time than expected by the person who stutters. He or she consequently has an anticipated auditory expectation that cannot be fulfilled and is in conflict with the actual time needed for the production of the vowel, leading to an incorrect interpretation of the articulatory process (Harrington, 1988). The person who stutters therefore assumes that the vowel was produced later than planned, although the opposite is the case: the vowel is expected too early, it is being anticipated. Explaining stuttering symptoms with this model, Harrington hypothesizes that a stuttered syllable is the result of the attempt to correct this presumed asynchrony. In order to correct this perceived asynchrony, the production of the preceding consonant has to be corrected to be realized at an adequate temporal distance from the vowel. Since this assumption is a misinterpretation of the person who stutters, meaning that the production of the vowel was on time and only due to the anticipation it was perceived as delayed, the production consequently takes place too early.

Hence, the model postulated by Harrington implies that asynchronies are a core characteristic of speech productions in PWS. Moreover, it presumes that complete prevocalic segments or parts of it (i.e. consonants) are repeated. What the model cannot explain is the fact that some consonants are repeated various times and some prolongations last for seconds. Although this model has never been proven empirically, it receives support from various sides: on the one hand, studies on the neurological groundings of stuttering have demonstrated aberrations in the auditory cortices of PWS, as explained in chapter 2.5.2 (Kell et al., 2009), on the other hand, observations and behavioral data on the speech production of PWS compared to fluently speaking persons in different contexts also indicate that this model could be a plausible explanation for some symptoms and phenomena observed in stuttering. The model mostly concerns observations regarding the temporal aspect of stuttering, like the rhythmical structuring of speech by chorus speech or metronome speech that have – as already explained before – a strongly fluency-enhancing impact on disfluent speech, since persons affected by stuttering can now rely on a constant rhythm to orientate to. This leads to a simplified process of speech production by reliable auditory feedback and hence easier motor planning (Andrews, Howie, Dozsa, & Guitar, 1982; Guitar, 2014; Natke & Alpermann, 2010). Etchell and colleagues also worked on a model to explain this phenomenon and stated that it is the external rhythm of a metronome or joint singing/speaking compensating for the lack of internal rhythmical timing (Etchell et al., 2014). A compensation is also observable on neurological levels: instead of using the neuronal networks for internal timing, secondary systems are being used, that rely on external temporal cues in the sequencing of speech movement (Etchell et al., 2014). Harrington (1988) explains the effect of masking (i.e. covering one's own speech by white noise) leading to a more fluent speech as follows: since a person who stutters cannot hear his or her own speech anymore when masked by a noise, the person affected can also make no prediction about the planned syllable and therefore there can be no misinterpretation of his or her speech signal. The problem of expecting the articulation of a sound or syllable has thus been eliminated (Harrington, 1988). Delayed auditory feedback is one possibility to imitate the effect of a mismatch in one's own auditory feedback and the expectance of articulatory results in PWNS. As the name already implies, it returns one's own speech production delayed leading to lagged perception in the speaker, which in turn causes interruptions of the onward flow of speech that are very similar to those observed in stuttering. By this means, a kind of artificial or externally triggered stuttering can be caused that might go back to the same mechanisms as developmental stuttering in Harrington's model. Of course, delayed auditory feedback can also be used in the opposite manner, i.e. as a fluency-enhancing condition for PWS (1988): for some persons affected by stuttering, it has been observed, that this delayed auditory feedback led to a more fluent articulation, probably due to the fact that now the mismatch in one's own auditory feedback and the expectance of articulatory results was eliminated (Harrington, 1988).

One very important component of rhythm and timing, which comprises processes of perception and production and is therefore a very important concept in this field, is so-called *sensorimotor synchronization*. This term shall be explained and put into context with stuttering. Its relevance in the research about the disorder shall be depicted in the following section, as well.

2.6.3.1 Sensorimotor synchronization

Before depicting the relevance of this concept in the context of stuttering research, a brief definition and explanation of it is needed to follow the hypotheses of chapters 4 and 5. Sensorimotor synchronization (SMS) is the rhythmic coordination of perception and action or simply the rhythmic reaction to an external rhythm (Repp, 2005; Repp & Su, 2013). It is present in various contexts: the most natural situation in which SMS is of great importance is playing within a musical ensemble or dancing to music. However, simple reactions to music, such as the clapping, stamping, or nodding to a beat, are also variants of SMS, irrespective of whether the given rhythm stems from a metronome, a musical excerpt or even speech. As the name already implies, there are two components of SMS. The term '*senso*' suggests that there are processes of perception, that are also known as *covert processes*. These are the basis for the generation of a rhythmic response, as the term *'motor'* suggests. This response falls under the name of *overt processes (*Repp & Su, 2013). Hence, SMS consists of two single processes, which only work together as a combination of both. This is because a covert process always comes prior to an overt process and SMS is only complete if both processes consolidate to the joint process of SMS. There are different options to synchronize to a given rhythm. Since a given rhythm usually depicts something constant, there is an inter-onset-interval (IOI) between those regularly occurring events. A metronome is a good example, as it always displays the same magnitude and, therefore, depicts an isochronous auditory sequence. When synchronizing to this rhythm, there are different ways to do so. The basis for any synchronization, however, is the recognition of this rhythm. The rhythmic reaction can be in-phase, meaning that the rhythmic response (i.e. movement) is taking place at the same time as the pacing event itsself. In case of anti-phase reaction, the movement takes place exactly in the middle of the IOI. Those two basic forms of synchronization can be varied in their rate: for example, two movements can coincide with one beat of the metronome or, in the opposite case, a rhythmic reaction might only be observed on every second beat of the metronome. Those variations are, of course, diverse and can also take place at faster or slower rates (Repp, 2005).

Most of the literature on SMS focuses on tasks like tapping or clapping to a beat. For the paper at hand, the chosen variant of SMS is a verbal response to a regularly occurring pacing event. Therefore, research literature on tapping tasks will not be explained in detail, only relevant information that also applies to verbal synchronization tasks will be presented. What comes first in any kind of synchronization is the intention to synchronize, since intentional movements are required in the overt process (Repp, 2010; Repp & Su, 2013). Besides the intention to synchronize to a given rhythm, the tempo of the pacing stimulus is also relevant to the feasibility of the task. Performance can vary significantly, depending on age and musical experience of participants. Therefore, there is not a clear cut-off value in verbal synchronization, since a clear maximum limit is given by biomechanical abilities. However, there seems to be a minimum level for synchronization that can also be applied in the verbal domain. This minimum level for synchronization does not depend on biomechanical abilities, but rather on the ability to foresee upcoming pacing events. Since those tasks require a very precise reaction which must be predicted and is not limited by articulatory movements, studies demonstrated that the value of 1.8 seconds can be taken as a cut-off for the IOI. Values exceeding those 1.8 seconds lead to a difficult and imprecise prediction of the upcoming pacing event. In such a case, the (verbal) reaction to a pacing event cannot be counted as a prediction of but only as a reaction to the pacing event (Repp, 2005, 2010).

In the context of SMS one phenomenon must be highlighted: the negative mean asynchrony (NMA). As the name already implies, negative mean asynchrony stands for the observed phenomenon that asynchronies, i.e. synchronization errors tend to occur some milliseconds prior to the actual pacing event and are therefore not distributed symmetrically around it. Those results were first found for tapping tasks but can also be applied to other forms of motor synchronization to a given rhythm (Aschersleben, 2002; Repp & Su, 2013; Woodrow, 1932). Despite many attempted explanations and extended research, this phenomenon has not yet been fully explained. What is clear, however, is the fact that NMA is not as prominent or even completely absent in musical contexts, which became apparent when testing musically-trained persons regarding this issue (Aschersleben, 2002). Furthermore, it became evident in a study by Aschersleben (2003) that most of the participants were not aware of this anticipation. Hence, subjective synchrony does not correspond to objective synchrony. On top of that, there is a lot of interindividual variability: whereas some participants anticipate up to 100 milliseconds, others barely show a NMA, with the latter mostly being observable in musically-trained persons (Aschersleben, 2002).

The ability of SMS develops over the course of many years: those are the results of a study by De Bruyn and colleagues (2008) which investigated children between three and eleven years of age concerning their development in tasks of SMS. However, synchronization was not tested in a verbal modality but by tapping to rhythm in this study. Since there are to date no studies focusing only on verbal synchronization development, those studies must be used instead for gaining information on the development of SMS. While the youngest participants did not adapt to the rhythm of the music but stayed within their self-chosen tempi, there was a growing effect of synchronization observable in five-year-olds. The ability to synchronize to a given rhythm improved constantly with age, with the biggest development taking place between three to seven years. Performance therefore reaches adult levels quite early, mostly around seven years of age (De Bruyn et al., 2008; Monier & Droit-Volet, 2019). When looking at the abilities of SMS in adults only, it was mostly the effect of musical training that was relevant for the ability to synchronize, since professional musicians showed a significantly reduced variability of inter-tap-intervals compared to untrained participants. This effect was, however, only observable in professional musicians, but not in amateurs (Repp, 2010).

2.6.3.2 The link between sensorimotor synchronization and cognitive parameters

After a basic yet comprehensive depiction of SMS, the following section shall explain why it is interesting and important to look at skills of SMS in different populations and what those performances of SMS can reveal about certain skills of children and adolescents. Before going into detail about the specific goals of testing SMS in a population of PWS, general information on links between skills of SMS and further cognitive parameters shall be given. In speech, parameters of time are essential to recognize boundaries between words, syllables, and segments within the speech flow. The perception of those cues is relevant even in the earliest stages of speech development in children. Neuronal encoding of speech as well as the synchronization to a rhythm require precision when it comes to processing temporal structures (Clark & Clark, 1977). This leads to two - at first sight - very distinct processes making use of one common and very basic skill.

The ability to read, as a very advanced skill of speech and language, was also linked to SMS. A poor reading ability has frequently been ascribed to decreased neuronal prediction, leading to rhythmical and phonological deficits (Woodruff Carr, White-Schwoch, Tierney, Strait, & Kraus, 2014). It is therefore assumed that the sensitivity for timing in speech plays a crucial role in language acquisition and boosts the processing of phonological skills. Research has demonstrated that a link exists between the ability to synchronize to a beat, which demands a fine-tuned auditory-motor coupling, and speech skills, not only in school-aged children but also in adults (Huss, Verney, Fosker, Mead, & Goswami, 2011; Thomson, Fryer, Maltby, & Goswami, 2006; Thomson & Goswami, 2008; Tierney & Kraus, 2013). A study investigating 35 children aged three to four years demonstrated again that those children, who can synchronize to a beat also showed a better performance in the neuronal processing of speech and therefore performed significantly better in tests of early speech and language skills. Authors therefore stated that the ability to neuronally encode temporal structures in speech is a basic competence for the acquisition of reading (Woodruff Carr et al., 2014). In sum, studies investigating the link between timing and reading skills show that children as well as adults with difficulties in reading or writing show impairments in various tasks of timing. This is typically demonstrated in tapping tasks, where a great variability in taps in combination with an enhanced tendency to anticipate stimuli is observed. Measurable aberrations seem to derive from aberrations in the perception of rhythmic stimuli or from an insufficient perception of the discrepancy between the own rhythmic response and the actual rhythmic stimulus (Birkett, 2014).

But it is not only the ability to read that seems to be linked to rhythm and timing. According to Tierney and Kraus (2014), phonological skills are also linked to skills in SMS. They postulated a model to expand on this idea and to investigate the link between those two skills, which is known as the *precise auditory timing hypothesis (PATH)*. In this model the authors pick up on the fact that musical training can improve phonological skills but also admit that, to date, it is unclear how this cross-domain enhancement works. They state that language (i.e. the detection of boundaries, turn-taking or vowel and consonant discrimination) and rhythmic skills depend on fine-grained details of sound regarding timing and therefore conclude that auditory-motor timing is a feature that demands "[…] the pre-conditions necessary for cross-domain enhancement to occur.“ (Tierney & Kraus, 2014, p. 1). Here, an overlap of two initially quite distinct abilities is assumed, which does not only concern the skills themselves but also is also to be found on neurological levels: not only the ability to synchronize to a certain rhythm but also the ability to recognize the rhythm of a certain language in form of stressed and unstressed syllables demand an exact perception of details in timing. On neurological levels, neuronal networks for the processing of temporal structures of speech and music also show connections and overlaps, as it was already demonstrated in chapter 2.6.2 (Tierney & Kraus, 2014). Those assumptions are based on results from a study in 2013 which demonstrated that participants with a lower variability in tapping also performed better in a non-word reading task (Tierney & Kraus, 2013). The assumption postulated by PATH therefore is that musical training enhances explicitly phonological skills due to an improved ability to make use of temporal cues regarding the duration while perceiving speech. Thus, the perception of speech timing is a basic and essential requirement for the acquisition of phonological skills with PATH creating a link between existing results in research. Furthermore, the authors suppose that not only reading abilities and phonological skills are influenced positively by musical training, but also other cognitive skills, such as executive functions (Tierney & Kraus, 2013, 2014; Woodruff Carr et al., 2014).

Additionally, other disorders or cognitive impairments were found to be linked to musical abilities: children affected by the attention-deficit/hyperactivity disorder (ADHD) and children with specific language impairment displayed a poorer performance regarding their abilities in perception and production of rhythm compared to healthy controls (Jentschke et al., 2008; Noreika, Falter, & Rubia, 2013; Sallat, 2011). Although those studies did not test SMS directly but rather basic musical skills, results still show that timing and musicality can be very sensitive indicators for various disorders. However, more research is needed to clarify the exact mechanisms and connections. It has been verified so far that SMS specifically, as well as timing generally, are linked to further cognitive functions, leading to aberrations within those skills in children and adults affected by ADHD, specific language impairment or dyslexia. According to those results, researchers even raise the question, whether atypical rhythm is "[…] a risk factor for developmental speech and language disorders[…]" (Ladány et al., 2020, p. 1).

2.6.3.3 Sensorimotor synchronization in stuttering

One of those disorders of speech and language – or rather of speech fluency – might be stuttering: as already mentioned above in chapter 2.6.3 it has been discussed for a long time that stuttering might be caused by or is at least associated with a timing deficit (Etchell et al., 2014; Falk et al., 2015; Ludlow & Loucks, 2003; Max et al., 2004; Max & Yudmann, 2003; Ning, Peng, Liu, & Yang, 2017; Olander et al., 2010; Sares, Deroche, Shiller, & Gracco, 2019). So far, timing abilities in PWS have been tested with some verbal and non-verbal motor tasks. Studies have demonstrated that PWS are generally less flexible and therefore adapt less efficiently to disturbances during speech production. Furthermore, their speech motor system seems to be more vulnerable and is more likely to be disturbed by increased demands like stress or a high linguistic level (Namasivayam & van Lieshout, 2011). As already shown in chapter 2.6.3, the exact reasons for the loss of motor control are not completely understood yet and one of the most popular hypotheses at the moment is the hypothesis of a dysfunctional timing mechanism within the motor system leading to a partial or even full loss of control during speech in stuttering (Boutsen, Brutten, & Watts, 2000; Harrington, 1988; Max & Yudmann, 2003; Olander et al., 2010).

Since the idea of stuttering being associated with a timing deficit has been specified before, the focus of the current chapter shall not be on general timing in stuttering but rather on SMS in stuttering. It was as early as 1967 that Herndon investigated the ability to differentiate the duration of two tones in PWS and PWNS (Herndon, 1967). This ability only represents the perceptive part of SMS; however, it contains very relevant information given the fact that most other studies on SMS exclusively focused on productive tasks of SMS. Herndon (1967) found that PWS performed significantly below their fluently speaking peers in the assessment of tone durations and therefore saw his hypothesis of problems with temporal cues as confirmed. Many further studies investigating the motor component were able to replicate results in the motor domain of SMS: the tempo of vocal or manual reactions was reduced, speech tempo was slower and the initiation of oral and manual movements was also partly delayed (Hulstijn et al., 1992;

Smits-Bandstra, De Nil, & Saint-Cyr, 2006; W. G. Webster, 1990). Of course, those results lead to an even larger interest in the causes of this phenomenon. The methods chosen to investigate SMS were mostly designed in a way that the motor component was tested mandatorily, since most tasks involved oral or manual reactions. Consequently, a precise differentiation between perceptive or productive components was not possible since a motor task always involves a preceding perception. This fact led many authors to focus on motor components when proposing models and theories. It is hypothesized that an elevated threshold for the neural activation of motor components is one of the reasons for the delayed manual movements (Alm, Karlsson, Sundberg, & Axelson, 2013; Sommer, Wischer, Tergau, & Paulus, 2003). Studies conducted with adults also show partly contradictory results: Max and Yudman (2003) tested ten PWS and ten fluently speaking persons on their SMS abilities. They used a paradigm in which participants heard stimuli with three different IOIs for various speech and non-speech tasks: the production of a syllable to a given rhythm but also the silent contact of lips or the contact of index finger and thumb to one of the given rhythms were supposed to cover the spectrum of speech, oral and other motor behavior. There was a synchronization and a continuation phase for all conditions. Hence, this study, too, predominantly focused on the motor component of SMS. Results show that both groups performed similarly regarding their timing variability, as well as their accuracy, displaying no significant differences in all three tasks tested and both phases. Subsequently, authors concluded that PWS do not differ from PWNS in terms of their ability to orally (speech and non-speech movements) or manually synchronize to a given rhythm, thus rejecting the theory of a deficit in the timing domain (Max & Yudmann, 2003). Since this study focused on adult PWS only, it is therefore not primarily relevant for the thesis at hand. Additionally, it must be stated that the number of participants was relatively low.

A very recent study, however, focused on the perceptive component of SMS in a population of children from six to eleven years comparing CWS and fluently speaking peers. The task chosen to test auditory discrimination of rhythms consisted of two simple and one complex rhythm. A certain rhythm was presented twice and followed by a third rhythm, which was either again the same or a new one. Children were asked to decide whether the third rhythm perceived was the same or different (Wieland et al., 2015). This way, the authors managed to focus on the purely perceptive part of SMS. Results of the study show clearly that CWS perform significantly worse in the discrimination of rhythms than children who speak fluently. Hence, first evidence of an impaired rhythm perception in CWS was provided. Another study, however, that also investigated the ability of SMS in CWS, found less clear results: Olander and colleagues tested

17 CWS aged four to six years and compared them to a fluently speaking control group on a synchronization-continuation-paradigm in the motor component (hand clapping). Results suggest that there is a subgroup of CWS that display a non-speech motor timing deficit (Olander et al., 2010). Since this study by Olander and colleagues used a paradigm involving motor skills, results cannot be compared to the one by Wieland et al., which might also explain why results are neither convincing nor unambiguous by comparison. Another study involving perceptive and productive components of SMS was conducted by Falk and colleagues: they investigated non-verbal timing skills in 20 children and adolescents who stutter and a fluently speaking control group (Falk et al., 2015). Participants were asked to synchronize to a given rhythm by finger tapping. Results showed that 65% of participants who stuttered showed a significantly worse performance in SMS, specifically in terms of accuracy or consistency or even both. It became evident that children with a weaker performance on SMS also displayed more severe stuttering symptoms. Hence, those results support the idea that children and adolescents who stutter display a timing deficit that becomes evident in tasks of SMS – in the combination of perceptive and motor tasks. Unfortunately, there was no task to also test the verbal domain. However, this study still provides very relevant information, since it also investigated the interesting population of children and adults who stutter and came to the conclusion that there seems to be a timing deficit as measured by tasks of SMS.

Those studies are discussed in detail in chapter 4. Despite some heterogenous results in this field, there is a clear tendency. Most studies can find aberrations in the timing domain, supporting the hypothesis of a SMS deficit in children and adolescents who stutter.

2.6.3.4 Summary of recent scientific results

Having introduced the role of perceiving rhythmical structures for speech and SMS as well as having presented the recent studies on SMS in the context of cognition and language, a summary on the current state of research in SMS within the context of stuttering shall be given to gain a framework of the relevance and importance of the research questions of the thesis at hand.

The fact that rhythmical elements are a very helpful tool to enhance fluency in the speech of PWS has been known for many years or even decades now (Van Riper, 1982, 1986). At the same time, it became evident quickly that those fluency enhancing techniques like paced speech do not lead to a stable and permanent therapeutic effect and were not suitable for the daily use. For those reasons, fluency enhancing techniques are barely used for therapeutic interventions but mostly in the context of research nowadays. However, what has been gained from these discoveries is the knowledge that there is a strong link between rhythmic abilities and developmental stuttering: Many studies, models and theories have tried to investigate and explain this issue since then. Besides Harrington’s model (1988), there are also newer models like the one by Max and colleagues (2004) which propose the assumption that PWS display deficient speech-specific feedback and feedforward processes. Other authors, however, do not necessarily see the emphasis of this disorder in an aberrant perception or a deficient process of speech processing, but rather within the motor components. They suppose it is a disorder of movement control that is causal for the phenomenon of stuttering and therefore raise the question whether stuttering is to be compared to other motor disorders such as spasmodic dysphonia or Tourette’s syndrome (Ludlow & Loucks, 2003). In addition to those two perspectives on stuttering, there is also a third one that can be found somewhere between those two extremes: Olander and colleagues (2010) are of the opinion that a motor timing deficit is of causal nature in stuttering, since results demonstrated that PWS displayed a more variable performance in clapping to a beat than PWNS. Furthermore, the authors concluded that there is at least one subtype of stuttering children that is affected by a non-speech motor timing deficit as they found out that there is a great overlap in speech and non-speech processes of motor control in the same time span when stuttering occurs and becomes chronic. This finding can provide interesting details about motor deficits observed in adult PWS regarding the question whether they are of causal or of compensatory nature (Olander et al., 2010). Additionally, other authors tried to investigate the link between rhythmic abilities and speech by testing rhythmical abilities in a speech context (Max & Yudmann, 2003). Unfortunately, the results are not conclusive in this domain, since some results did not show significant differences between stuttering and non-stuttering participants, which is in contrast to other studies named before.

Many neuroimaging results, however, indicate that areas relevant for the perception and production of rhythmic elements, show aberrations in PWS. Unfortunately, results on neuroimaging in stuttering cannot answer the question if the observed rhythmic-motor processes are causally linked to developmental stuttering. The results that have been collected so far might imply, though, that there exists a strong link between rhythm perception and production and stuttering.

The following three chapters of this thesis are trying to add new information to those questions by investigating different aspects of rhythm and stuttering in a population of children and adolescents affected by stuttering. First, speech and articulation rate shall be investigated as a very basic measurement of rhythm and timing in a natural setting. Second, SMS skills are investigated more closely by means of a task on verbal synchronization, as this field explicitly is still underrepresented when it comes to SMS tasks because most studies focused on non-verbal motor components and, on top of that, most research was conducted on adults. Third, the question is posed whether rhythmic skills can predict stuttering severity after therapy and, therefore, the success of a stuttering therapy. The fifth chapter, in particular, aims to give new and relevant insights in the treatment of stuttering and the potential power of SMS skills to be used in the diagnostic and therapeutic context of stuttering (Ladány et al., 2020).

3. Speech rate and articulation rate in stuttering

In the context of timing and rhythm, it might seem surprising at first to discuss speech rate and articulation rate in PWS. However, speech rate[3] and articulation rate are important global measures reflecting verbal output (Pellowski, 2010). Whereas speech and its synchronization to rhythms will be investigated in the chapter to follow, a rather natural setting for analyzing speech and articulation was chosen for this first study. The analysis of speech and articulation rate can add valuable information to this topic, since for this purpose speech data can be collected in a more natural setting than is possible for rhythmical tasks. Furthermore, two different modalities were chosen to collect data: Reading and speaking. The comparison of these two modalities adds interesting insights, especially when comparing the two groups.

Speech is, as discussed previously in chapter 2.6, a rhythmic concept per se, since temporal regularities "are present at multiple hierarchical levels […]" (Ladány et al., 2020, p. 4). Even if in speech those regularities sometimes are harder to perceive due to a higher variability caused by quasi-periodic intervals (compared to music, where intervals are periodic and rhythm, therefore, is more salient), spoken language entails rhythm. This rhythm is carried by the amplitude envelope, as explained before (see chapter 2.6.1). Comparable to musical rhythm, speech shows grouping of weak and strong events that form metrical structures: the pattern of unstressed and stressed syllables. Those patterns play a crucial role in speech processing, as discussed before, but also in language acquisition (Ladány et al., 2020).

In stuttering, specifically, speech rate and articulation rate are considered to play important roles not only in onset and development, but also in the persistence of stuttering. Speech rate is, furthermore, considered an essential part of the diagnostic processes in PWS (Erdemir, Walden, Jefferson, Choi, & Jones, 2018; Pellowski, 2010) and the reduction of speech rate is part of established therapeutic interventions in stuttering, like, for instance, prolonged speech (see chapter 2.4.4). Speech and articulation rate, with their many relations to interesting phenomena known to appear in stuttering, therefore seem a worthwhile topic of investigation.

3.1 Introduction

Speech rate is usually defined as the total number of units (syllables or words) divided by the time needed to produce the speech sample e.g. syllables or words per minute/second (Pellowski,

---

[3] Sometimes also the term speaking rate is used in the literature to refer to the same construct; for the paper at hand the term speech rate will be used, analogously to current literature on this topic.

2010). Speech rate is a very traditional clinical measurement that usually entails all kinds of pauses and disfluencies (Chon, Sawyer, & Ambrose, 2012; Davidow, 2014; Hall, Amir, & Yairi, 1999). It is supposed to describe verbal output rather globally; the precise timing of articulatory gestures, however, is not depicted by this term. Early research on this topic within PWS showed mostly slower speech rate due to interruptions caused by disfluencies compared to fluently speaking persons, with rates of about 122,7 words per minute in PWS compared to 170 words per minute in fluently speaking persons (Bloodstein, 1944). Speech rate can not only be used to diagnose stuttering severity; during the years of research on this topic other questions like the influence of parental speech rate on the development of stuttering, or the assessment of therapy outcome were investigated: For example, it has been reported that mothers of CWS showed a significantly higher speech rate when speaking to both CWS and CWNS (children who do not stutter), in comparison to mothers of CWNS (Meyers & Freeman, 1985). Concerning therapy outcome measures, an approximation of speech rate in PWS to rates of PWNS was expected as a result of enhanced fluency, also leading to more speech naturalness in listener-judged experiments (Hall et al., 1999; Ingham, Martin, Haroldson, Onslow, & Leney, 1985).

For many researchers, however, articulation rate is the more precise way to describe the speech motor execution or the motor transition ability, since it reflects the speaker's ability to coordinate respiratory, phonatory and articulatory processes (Tumanova, Zebrowski, Throneburg, & Kulak Kayikci, 2011). There is no bias caused by disfluencies slowing down speech rate and, therefore, articulation rate seems like a more precise estimate of time needed for speech motor execution. Articulation rate is defined as the number of perceptually fluent syllables in each utterance divided by the duration (often: seconds) of the utterance after removing all instances of disfluencies and pauses greater than 250 milliseconds (Chon et al., 2012). A slow articulation rate may, therefore, signify an immature or compromised speech motor control system (Erdemir et al., 2018; Pellowski, 2010).

Two major hypotheses on the link between speech rate/articulation rate and stuttering were postulated: First, psycholinguistic models (see chapter 2.5.3) have explained the observed slower speech rates in PWS with additional time needed to process linguistic and phonological information and to plan the following speech movements. It has also been discussed that this lower speech rate might derive from compensatory strategies to avoid stuttering symptoms, such as the planning of rephrasing (Peters, Hulstijn, & Starkweather, 1989). In support of this hypothesis, Meyers and Freeman (1985) found slower speech rates in the perceptually fluent speech of CWS compared to fluently speaking peers. In detail, they found this effect to be even stronger in CWS with more severe symptoms. The second hypothesis is based on the exact opposite idea and states that CWS speak at a rate that exceeds their speech-motor control capacities or language abilities and therefore increase their chances of disfluencies – in other words, CWS articulate faster than they are able to generate correct phonological encoding (Hall et al., 1999). This idea is in line with the advice often given to children affected by stuttering– to speak at a slower rate. Indeed, the articulation rate of children who were later diagnosed with stuttering was found to be faster than that of children who spoke fluently and remained fluent (Kloth, Janssen, Kraaiamaat, & Brutten, 1995). However, research findings on speech rate and articulation rate, especially within the population of PWS, have been inconclusive due to various factors: Research methods vary greatly in the handling and definition of excluded segments like pauses and disfluencies. Furthermore, there is no uniformity in the measure chosen (syllables per second, words per minute, etc.) and the types of utterances investigated (all utterances versus only perceptually fluent ones). On top of that, samples are very heterogenous concerning the age of the population investigated, the time since stuttering onset, stuttering severity and sample size (Erdemir et al., 2018; Sawyer, Chon, & Ambrose, 2008).

To date, two longitudinal designs have tried to clarify the link between speech and articulation rate and persistent or recovered stuttering: In a comparison of perceptually fluent utterances of CWS Hall and colleagues (1999) found that CWS tended to show slower articulation rates compared to fluently speaking peers. When comparing persistent and recovered CWS on articulation rate, results failed to reach significance. However, they showed a clear tendency for children who later recovered from stuttering to display slower rates than persisting children (Hall et al., 1999). The authors therefore suggested to use articulation rate as a kind of prognostic indicator for the two possible divergent directions: stuttering versus recovered. It is important to note that results failed to reach significance between groups when using syllables per second to measure articulation rate but reached significance between groups when using phones per second. This fact makes results harder to compare to the study at hand or to other studies on this topic, since in most other studies syllables per seconds is used to determine articulation rate. On top of that, rates should not point towards different results only because of changing the units used. Another investigation by Kloth et al. (1999) examined fluent utterances of children at two distinct points: before stuttering onset and then again, one year after onset. They found a higher variability concerning the articulation rate in persisting children than in recovered children – at both moments of investigation (Kloth, Kraaiamaat, Janssen, & Brutten, 1999).

In their investigation of speech and articulation rate in school-aged children Logan et. al (2011) chose three different tasks (sentence priming, structured conversation and a narration task) to compare rates between CWS and CWNS. Results concerning speech rates show a higher speech rate in CWNS compared to CWS and within the group of CWS speech rate was negatively correlated with stuttering severity. However, when comparing articulation rate, the authors did not find significant differences between CWS and CWNS, indicating that perceptually fluent speech does not differ between groups (Logan et al., 2011). Similar results were obtained in a study by Usler and Walsh (2018) using 4 sentences with different syntactic complexity. Also using syllables per seconds, they could not find significant group differences in school-aged children (Usler & Walsh, 2018). A further study interested in the link between stuttering severity and articulation rate came to differing results as they found out that a slower articulation rate in CWS was associated with a higher frequency in stuttering-like disfluencies, and longer durations of prolongations, therefore indicating a significant correlation of these constructs (Tumanova et al., 2011). Note that this study was conducted with pre-school-age children. Investigating articulation rate in a comparable age-span, Erdemir et al (2018) conducted a study with children from three to five years. He subdivided them into groups of persisting, recovered and nonstuttering children and added the influence of emotion to his investigation of articulation rate. Results indicate that only within the group of persisting CWS did the negative emotion condition lead to a significantly slower articulation rate. Within the groups of recovered or nonstuttering children, no effect of emotion was measurable. On top of that, faster articulation rates within fluent speech were found in recovered compared to persisting children. Those results hint towards a rather instable and immature speech-motor system in persisting CWS compared to children who recover and suggest interactions between emotional processes and speech-motor control as hypothesized in chapter 2.5.2.1, where studies found an incomplete myelinization in the fiber tracts of FLS and AF that are close to regions of emotional processing. Erdemir and colleagues (2018) therefore hypothesized that emotionally complex situations might lead to interferences of activation in these closely located areas leading to stuttering symptoms caused by a disturbed information transport (Johnson et al., 2010). Authors therefore concluded that CWNS and recovered children seem to rely on more stable and mature systems of speech-motor control. In CWS, however, the comparison of articulation rate in fluent versus stuttered speech after negative emotions could be used to predict persistency of or recovery from stuttering (Erdemir et al., 2018).

In sum, these investigations represent a very inhomogeneous picture of speech and articulation rate in PWS, which is a further reason for the thesis at hand to specifically investigate those constructs within this population. Speech and articulation rate can be measured in various settings, as demonstrated above. Speech samples can be elicited in many different ways, which again makes it harder to compare results with each other: Whereas spontaneous speech samples or unstructured conversations (Hall et al., 1999) represent a very natural setting, sentence priming or narration tasks (Logan et al., 2011) add additional demands on speech production. Also, reading aloud represents a setting with high demands on all speech related systems, especially within children and adolescents who are still in the process of building this competence and the population of PWS who are confronted with frequent disruptions of fluent speech. Therefore, analyzing speech and articulation rate when reading aloud might add relevant information to the comparison PWS and PWNS and of course, to the comparison of those two settings – a very natural conversation and a rather challenging task like reading aloud. Since reading represents a task with its very specific challenges and it, furthermore, underlies changes within the natural development, a brief introduction to reading development in general and specifically in PWS will be given.

Reading aloud is a complex competence that develops over many years beginning in early school years and lasts into adulthood. It requires various cognitive capacities, such as working memory and attention, but also literacy skills, such as phonological and orthographic awareness, morphosyntactic knowledge and vocabulary, all leading to the process of comprehension (Kim, 2015). In reading aloud, speech motor skills are required, adding further demands on the reader, especially in the case of CWS. Reading fluency consists not only of fluent articulation, but also of the preceding process of text decoding. And only if those two processes run successfully can a prosodic performance ensue that entails a meaningful structuring of the text (Franke, Hoole, Schreier, & Falk, 2021; Rasinski, 2004). Fluent articulation as one major component of reading aloud can be a very challenging task, especially for children and adolescents who stutter and who might be - on top of that - still in the process of reading development. However, reading ability in PWS (of any age) is a field with many inconclusive results, due to factors such as: different age groups investigated, different tasks used, different modes of reading (silent or aloud) and different definitions of successful reading (i.e. comprehension versus fluency).

The development of reading fluency is composed of different skills: One of them is the so called *decoding ability* that is usually developing to the stage of a decoding reader between 7 to 9 years, meaning that children this age are still in the stage of semi-fluency, unable to fluently read unknown words. Between 9-15 years they reach the level of comprehending and fluent readers, which is also the age-span of most participants for the study at hand. After 16 years usually the level of expert readers is reached (Landerl & Wimmer, 2008; Wolf, 2008). Reading rate strongly depends on age: It increases until young adulthood and starts to decrease in the forties (A. H. Chen, Khalid, & Buari, 2019). According to A. H. Chen et al. (2019) the average reading rate of 8-12 year old children is 144 words per minute (wpm), in teenagers (aged 13-19 years) it increases to 190 wpm (A. H. Chen et al., 2019). Those age groups reflect the age of participants in the study at hand, however, those results cannot be generalized, since they are derived from only one study (A. H. Chen et al., 2019). In reading accuracy, it seems that the development takes place up until 11-13 years; afterwards no significant improvement is found (A. H. Chen et al., 2019; Wolf, 2008). Further components in the development of reading fluency are the placement of respiratory pauses and the resulting prosody: The correct usage of respiratory pauses to structure the text, mark stress, divide sentences into smaller units (phrases) to generate meaningful chunks is something that has to be learned and is more likely to take place as soon as reading is getting more automatized and breath pauses are not distributed randomly any longer, but in a controlled and meaningful way. The competent use of pause patterns in a meaningful way to support prosody increases until the age of 13 years, when an adult level is reached (Godde, Bailly, & Bosse, 2022). In sum it has become clear that the age span in which reading fluency is achieved overlaps with the age of the younger participants of the study at hand. This factor must be kept in mind when interpreting results on reading fluency later.

It has been demonstrated above that results on articulation rate in non-read speech are rather inconclusive; when it comes to articulation rate in reading however, results show tendencies for slower rates: A study by Janssen and colleagues (1983) investigated reading rates in elementary school-aged children by analyzing a one-minute reading task and found that CWS produced significantly fewer words than CWNS. A further investigation amongst school-aged children (8-11 years) came to similar results when comparing oral reading rates (measured as fluent syllables per minute): Rates were significantly slower within the group of CWS compared to CWNS (J. S. Pinto, Picoloto, Capellini, Palharini, & Canhetti de Oliveira, 2021). Within the adult population (17-59 years), speech rate also was shown to be slower in PWS compared to PWNS in an oral reading task (J. Pinto, Schiefer, & Ávila, 2013). In an investigation of fluent read speech, Bosshardt (1990) also demonstrated slower articulation rates, not only in children (6 -10 years) but also in adults. One very recent study investigating articulation rate in a reading task (a one minute excerpt of a children’s book read aloud), however, did not find differences in articulation rates (Franke et al., 2021). Comparing all those different results, it is important to note that depending on data procedure, results can differ a lot: Whereas Janssen and colleagues (1983) only counted words per minute without further details about the handling of stuttering symptoms during the task, Franke et al. (2021) strictly excluded all kinds of disfluencies (stuttering like and other disfluencies) and on top of that also pauses that were caused by those disfluencies. Hence, the basis for statistical analyses on comparable questions differed strongly. The heterogeneity in the past results reflects the diversity in stuttering and its symptoms, but also suggests the need for further investigations (Nippold & Schwarz, 1990; J. S. Pinto et al., 2021). For the research question of the thesis at hand the focus lies strictly on reading fluency as measured in speech and articulation rate, but not on prosodic performance and comprehension.

After summing up all results of studies on speech and articulation rate in PWS, the goal of the thesis at hand is to add valuable information on this complex and multi-layered topic. In line with the tendencies shown in the literature so far, the following hypotheses are postulated:

- PWS are expected to display slower speaking rates due to stuttering symptoms,
- articulation rates however are expected to be comparable to PWNS after removing all disfluencies from the analyses.
- Severe stuttering is expected to have stronger influence on rates leading to slower rates and
- the influence of age on both speaking rates and articulation rates might be visible especially within the reading task.

3.2 Material and methods

3.2.1 Participants

54 native German-speaking children and adolescents displaying developmental stuttering participated in the experiment. The final sample of stuttering speakers consisted of 43 participants (5 females, 38 males) with developmental stuttering (mean age *M* = 12.83 years; *SD* = 2.46). Due to comorbidities (i.e. ADHD, Cluttering) or cognitive impairment, eleven participants had to be excluded from the analyses. Especially the comorbidity of cluttering has a strong impact on speech and articulation rate, which is why 3 participants with cluttering had to be excluded from analyses on speech and articulation rate (Canhetti de Oliveira, Broglio, Bernandes, & Capellini, 2013). A randomly selected control group was matched in age and gender (43 children and adolescents, 5 females, 38 males, mean age *M* = 12.74 years, *SD* = 2.43). Participants who stutter are in the following chapter abbreviated as PWS, to avoid

confusion, since CWS would not cover the age span of all participants and lead to confusion when talking about age groups. PWS were recruited and tested prior to a therapy course held near Munich during the summer of 2017, 2018, 2019 and 2020 (staerker-als-stottern.de). To avoid effects of therapy, the testing was done strictly prior to therapy. Participants of the control group were recruited through schools. Stuttering severity was assessed with the SSI-3 (Riley, 1994) and the German Version of the OASES (Yaruss & Quesal, 2006), with the latter being an assessment of the subjective psychosocial impact of stuttering on the everyday life of the participants. The SSI-3 scores ranged from mild to very severe stuttering, the scores for the psychosocial impact from very mild to severe.

Stuttering	**SSI-3**		**OASES**	
severity	**n**	**%**	**n**	**%**
1	2	4.7	1	2.3
2	10	23.3	16	37.2
3	11	25.6	22	51.2
4	11	25.6	4	9.3
5	9	20.9	0	0

Table 3.1 Distribution of participants per severity category for SSI-3 and OASES (*n* = 43)
SSI-3: 1 = very mild; 2 = mild; 3 = moderate, 4 = severe; 5 = very severe
OASES: 1 = mild; 2 = mild-moderate; 3 = moderate; 4 = moderate-severe; 5 = severe

In order to see how comparable groups are concerning two relevant cognitive parameters, namely working memory and inhibitory control, participants were assigned to two quick testing sessions prior to the interview and reading session and any therapeutic intervention: In the digit span, a subtest of the Wechsler Intelligence Scale for Children (Wechsler, 2010), PWS achieved a mean of $M = 9.49$ ($SD = 3.04$) and showed a performance comparable to the age-matched control ($M = 9.56$, $SD = 2.00$). Furthermore they participated in a Go-Nogo Paradigm, a subtest taken from the TAP-Battery (P. Zimmermann & Fimm, 2002), where results also showed a comparable performance regarding the error-percentiles, which are a good way of measuring inhibitory control. The group of PWS had a mean of $M = 51.30$ error percentiles ($SD = 31.68$) on this task and with a mean of $M = 54.14$ ($SD = 30.32$) the control group performed similarly. No significant differences between groups on any of these measures could be found (all $p > .05$).

In addition to the comparison of PWS and PWNS, age groups were formed to also allow for analyses between children (9-13.25 years, “young”) and adolescents (13.75 -17 years, “old”). There were 24 PWS (3 female, 21 male) and PWNS in the young group and 19 (2 female, 17 male) in the old group and same for the control participants. Concerning the parameters mentioned above (working memory and inhibitory control) homogeneity of variances was given and no significant differences between age groups could be found, indicating no influence of age on these measures (all $p > .05$). This information is relevant when comparing performances on speech and articulation rates, since all potential differences found are to be linked to age – especially in the case of reading - but not to general differences on cognitive measures between age groups. Furthermore, stuttering severity and the impact of stuttering displayed homogeneity of variances as well and showed no significant differences between age groups (all $p > .05$). Again, this fact must be kept in mind when interpreting results on rates, since any found differences in speech and articulation rates are merely to be ascribed to age and not differences in stuttering severity. Since no significant differences between the two age groups were found regarding any of the variables tested, the two age groups can be merged or analyzed separately, depending on the questions raised.

Participants and their parents were informed prior to the study and gave informed consent. The study was, of course, approved for conformity with ethical standards by the ethics committee of the faculty of medicine of the Ludwig-Maximilians-University Munich.

<table>
<tr><th></th><th colspan="3">PWS</th><th colspan="3">PWNS</th></tr>
<tr><th></th><th colspan="2">mean</th><th>SD</th><th colspan="2">mean</th><th>SD</th></tr>
<tr><td>Age
(in years)</td><td colspan="2">12.83</td><td>2.46</td><td colspan="2">12.74</td><td>2.43</td></tr>
<tr><td>Inhibitory control
(Error Percentiles of Go-Nogo task, TAP Battery, P. Zimmermann & Fimm, 2002)</td><td colspan="2">51.30</td><td>31.68</td><td colspan="2">54.14</td><td>30.32</td></tr>
<tr><td>Working memory
(digit span, Wechsler, 2010)</td><td colspan="2">9.49</td><td>3.04</td><td colspan="2">9.56</td><td>2.00</td></tr>
<tr><td>Education
(school type)</td><td>School type</td><td>n</td><td>%</td><td>School type</td><td>n</td><td>%</td></tr>
<tr><td>1= Primary School</td><td>1</td><td>13</td><td>30.2</td><td>1</td><td>12</td><td>27.9</td></tr>
<tr><td>2= Intermediate period (HS)</td><td>2</td><td>7</td><td>16.3</td><td>2</td><td>18</td><td>41.9</td></tr>
<tr><td>3= Intermediate period (RS)</td><td>3</td><td>9</td><td>20.9</td><td>3</td><td>8</td><td>18.7</td></tr>
<tr><td>4= Grammar School</td><td>4</td><td>14</td><td>32.5</td><td>4</td><td>5</td><td>11.6</td></tr>
<tr><td>Sex</td><td colspan="3">female: n = 5
male: n = 38</td><td colspan="3">female: n =5
male: n = 38</td></tr>
</table>

Table 3.2 Mean values and SD of PWS (*n* = 43) and PWNS (*n* = 43) on general measures

3.2.2 Stimuli and procedure

To collect speech samples, the participating children and adolescents were submitted to an interview and reading session, lasting about 10 minutes, depending on reading abilites and severity of stuttering symptoms. Participants were asked to sit comfortably at a table where they were recorded with a ZOOM H4N recorder (44.1kHz, 16 bit). An external headset microphone (beyerdynamic opus 54.16/3) was used for optimal acoustic quality; recordings were done in a quiet room with the experimenter present during the whole session.

For the interview, participants were asked comparable questions about their daily life and hobbies. The interview was supposed to last about 3 minutes at least to ensure the speech sample was sufficient and contained at least 60 seconds recordings of the participant talking. Afterwards participants were asked to read an excerpt from a German children's book (see Appendix 1) aloud, that is popular within the age range tested and recommended for readers from 8 years on (Maar, 2008). To ensure a good readability the excerpt was printed on two DIN

A4 pages in size 12.5 (Times New Roman). The selected excerpt combined narrative passages and passages of direct speech.

Note that there were 11 participants within the group of PWS that could not participate in this testing session. In this case, speech samples of the diagnostic session also consisting of an informal conversation about daily life and a read excerpt consisting of 250 syllables (on the topic of holidays) was used.

3.2.3 Data preparation and analyses

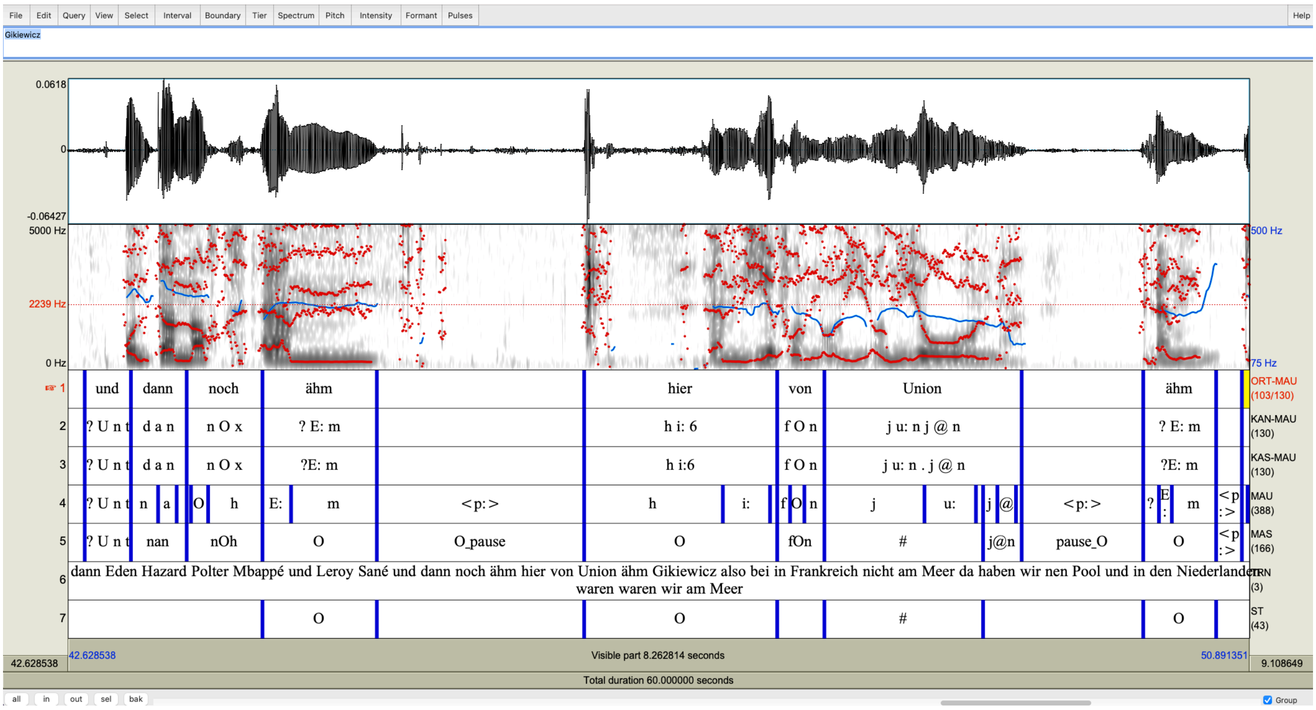
Before the main analysis all speech-data were edited with the freeware Audacity (Version 2.0.6) and Praat (Boersma & Weenink, 2009).

Data of the “interview” and the reading task had to be cut to an excerpt of 60 seconds each, which was done as follows: For the interview, passages of the investigator had to be excluded to only get speech material of the participant. With Praat (Boersma & Weenink, 2009), all those passages not containing speech of the participant were excluded and the rest of the participants speech was combined until 60 seconds of pure speech by the participant was reached. This file was then used for further annotations during the process of analyzing the interview data. For audio files of the reading task, procedures differed slightly: Since no interruptions of the investigator had to be excluded, the passage used for analyses was aimed to be the same for all participants. Starting point was not the beginning of the text, but the start of a paragraph within the first third of the text to give participants time to get into the reading and avoid disfluencies that might have been caused by excitement or discomfort caused by the testing situation. From this specified starting point, 60 seconds were added.

After cutting the excerpts (both – the interview and the reading task) to 60 seconds each, orthographical transcriptions had to be compiled. This was done manually to assure correctness and to account for variations caused by dialect or colloquial speech. With the audio file and the matching transcription, the segmentation into syllables could be done with the help of the “Pipeline without Automatic Speech Recognition”, an automatic segmentation software (MAUS = Munich Automatic Segmentation), a tool from the Bavarian Archive for Speech Signals (Kisler, Reichel, & Schiel, 2017; Schiel, 1999). With those pre-segmented text files, further annotation was implemented. Since speech rate is defined as the number of segments per time unit no further annotation was needed – disfluencies, pauses and stuttering symptoms do not have to be excluded here.

$$Speech\ rate = \frac{syllables}{seconds}$$

Articulation rate, however, excludes disfluencies of any kind – both stuttering and non-stuttering-like disfluencies; thus, all text files had to be edited to exclude segments that were affected by stuttering or other disfluencies like interjections or repetitions. For this purpose, an additional tier (see line 7, ST-tier in figure 3.1) was added where all segments affected were marked as # for stuttering and as O for other disfluencies, additionally those syllables were deleted from the syllable tier (see line 5, MAS-tier in figure 3.1), to not further include those syllables for the calculation of articulation rate. Furthermore, all pauses in direct proximity to disfluencies of any kind were analyzed regarding their cause: If they were directly linked to a stuttering specific cause (i.e. silent block etc.) or any other kind of disfluency (i.e. pause due to misreading/mispronunciation etc.) they were excluded as well using the markings #_pause or O_pause. To illustrate those markings, an example of a textgrid is shown in Figure 3.1.



6 dann Eden Hazard Polter Mbappé und Leroy Sané und dann noch ähm hier von Union ähm Gikiewicz also bei in Frankreich nicht am Meer da haben wir nen Pool und in den Niederlanden waren waren wir am Meer

Figure 3.1 Example of a Praat-file displaying the relevant markings to generate articulation rate

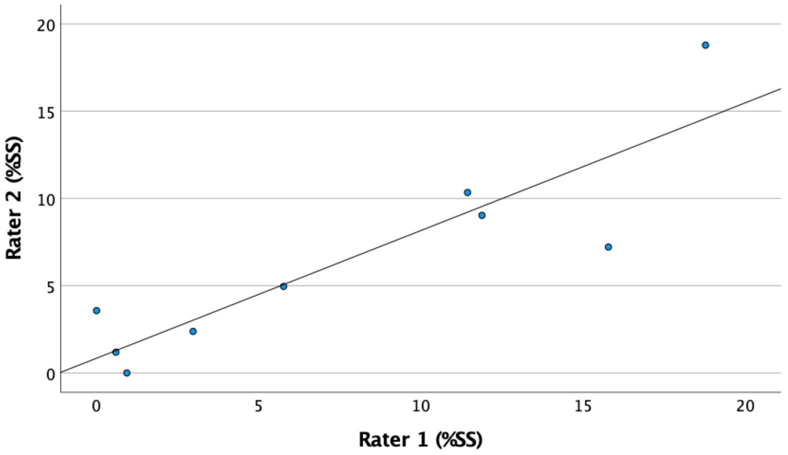
Since the calculation of articulation rate is a little more complex than speech rate, the formula will be given here for a better understanding:

$$Articulation\ rate = \frac{number\ of\ fluent\ syllables}{total\ duration\ of\ excerpt - duration\ of\ disfluencies\ (syllables + pauses) - pauses > 250ms}$$

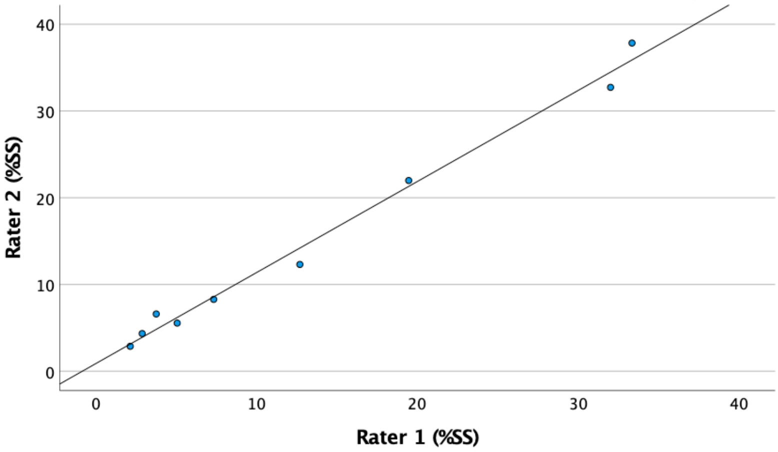
Since stuttering symptoms are not always easy to classify, especially when also marking all instances of other disfluencies, those annotations were only done by a trained speech therapist specialized on stuttering and in addition 20% of all files (spontaneous speech and reading task) were inter-rated by a second, independently judging research assistant, a speech therapy student, also specialized in stuttering. The amount of syllables excluded due to stuttering symptoms was calculated as the percentage of syllables stuttered (%SS) in relation to all syllables uttered - a commonly used measure of stuttering frequency also used in diagnostic tools (Riley, 1994). Results of both raters were then correlated to evaluate the agreement.

$$\%SS = \frac{syllables\ stuttered}{all\ syllables\ uttered}\ x\ 100$$

Results of Spearman-rho correlations between the %SS of both raters show very high levels of accordance for both tasks, in spontaneous speech a strong correlation with $r_s = .817$, $p = .007$ and in reading the correlation is even stronger ($r_s = .983$, $p < .001$). Results are visualized in figure 3.2a and b.



Correlation of Rater 1 & 2 on disfluent syllables in spontaneous speech



Correlation of Rater 1 & 2 on disfluent syllables in reading

Figure 3.2a and b Correlation of Rater 1 and Rater 2 for the %SS in spontaneous speech and reading

3.3 Results

In the following section results on rates in both conditions will be presented, starting with speech rate and articulation rate in the speaking condition, followed by speech rate and articulation rate in the reading condition and a final comparison of the two conditions.

3.3.1 Speech rate and articulation rate in speaking

The first part of this chapter reports results on comparisons between groups (PWS and PWNS, young and old) on rates during spontaneous speaking in the interview task. The second part focuses on possible causes for the differences found between groups. All statistical analyses were conducted in SPSS (Version 27). Normal distribution of the data was controlled for using the Shapiro-Wilk test, showing a normal distribution on speech and articulation rate ($p > .05$) between groups; between age groups however, in case of speech rate in older participants data lacked normal distribution slightly ($p = 0.15$). One outlier had to be removed for articulation rate due to extreme values (P28). The homogeneity of variances was checked as well, using Levene's test prior to further analyses, which reached significance for speech rate and for articulation rate (all $p < .05$), indicating no homogenous distribution of variances. The two-way ANOVA (type III sums of square; for the dependent variables speech rate and articulation rate with the between-subject factors group and age group) was chosen despite the lacking homogeneity of variances since they offered the best way of comparing groups in the given design (stuttering vs control; young vs old); on top of that, the variability of older PWS as explained later can be most likely held responsible for the lacking homogeneity of variances. After group comparisons, Spearman-rho correlations were performed to get a better understanding of factors influencing rates.

Analyses of speech rate demonstrated what had been hypothesized already: Speech rate is higher in PWNS than in PWS ($F(1,86) = 40.592$, $p <.001$, $\eta^2 = .331$) with an average rate of $M = 3.52$ syllables per second ($SD = 0.65$) in PWNS compared to $M = 2.53$ syllables per second ($SD = 0.92$) in PWS. Age groups did show slightly significant differences between younger and older participants on speech rate analyses ($F(1,86) = 3.979$, $p <.049$, $\eta^2 = .046$), with slower rates in younger participants ($M = 2.88$ syllables per second, $SD = 0.64$) than older participants ($M = 3.21$ syllables per second, $SD = 1.19$), which is also a result to be expected. A significant interaction of group and age group was found with ($F(1,86) = 6.840$, $p = .011$, $\eta^2 = .077$). Those results are visualized in figure 3.3: It becomes evident, that – besides the fact that PWNS show higher rates than PWS and older participants display higher rates compared to younger ones –

those results might also be carried by the fact that older PWS show an extremely high variability.

To further investigate this issue, t-tests were conducted comparing young PWS and PWNS on speech rate in speaking, displaying significant results with *t*(46) = -3.756, *p* < .001 with slower rates in young PWS (*M* = 2.57 syllables per second, *SD* = 0.65) compared to young PWNS (*M* = 3.18 syllables per second, *SD* = 0.47). Results on old PWS versus old PWNS with *t*(26.616) = -4 .824, *p* < .001 show slower rates but higher variability in old PWS (*M* = 2.46 syllables per second, *SD* = 1.19) compared to old PWNS (*M* = 3.94 syllables per second, *SD* = 0.60). Comparing young and old PWNS with each other, results also show significant differences with *t*(41) = - 4.628, *p* < .001 and slower rates young PWNS (*M* = 3.18 syllables per second, *SD* = 0.47) compared to old PWNS (*M* = 3.94 syllables per second, *SD* = 0.60). Looking at young and old PWS, though, results of the interaction found in the ANOVA and visualized in figure 3.3, are supported once again with no significant results of the t-test, but a huge variability in rates of old PWS (*M* = 2.46 syllables per second and *SD* = 1.19) compared to young PWS displaying a mean of *M* = 2.57 syllables per second with *SD* = 0.65. The effect of rising speech rates with age could therefore not be found within PWS – other than in PWNS, where a clear advantage of older participants was evident for the speaking condition.

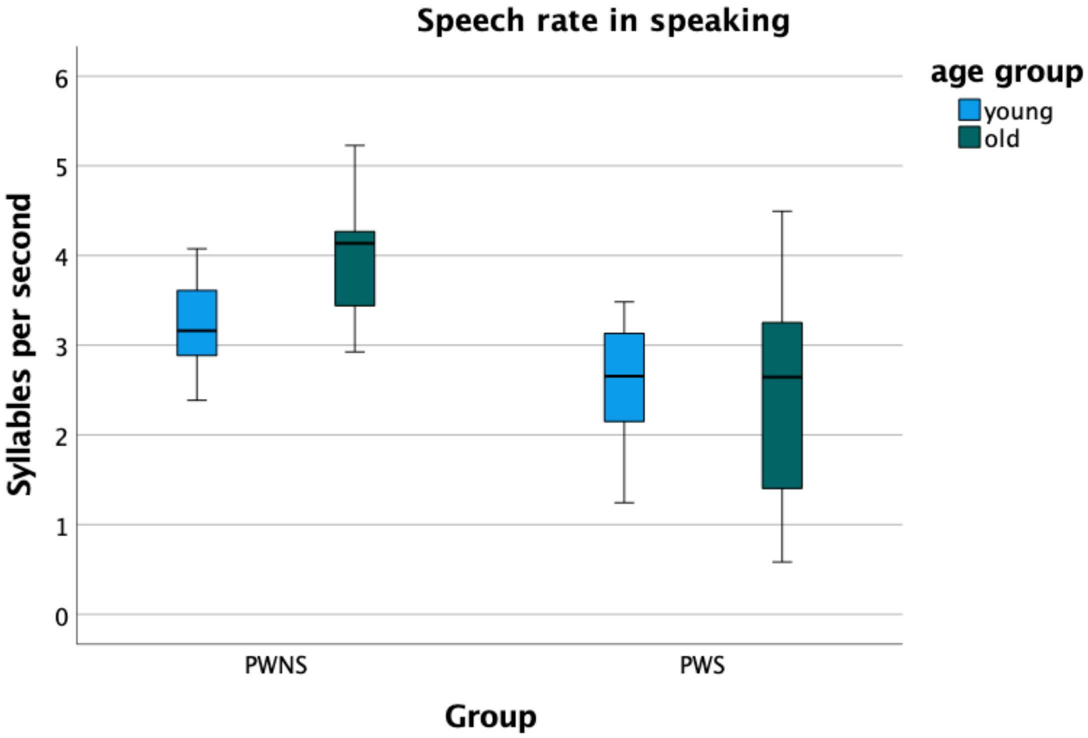


Figure 3.3 Speech rate in PWNS and PWS in spontaneous speech (interview task).
Bars represent speech rate (in both groups subdivided for age groups) from the first to the third quartile via the median. Whiskers ranging from minimum to maximum.

These results are most likely caused by more severe stuttering symptoms within older participants leading to a lower speech rate within this subgroup. The interaction of group and age group was therefore dissolved as it is evident within figure 3.3: In PWS – in contrast to PWNS – older participants to not display the significant rise in rate.

To illustrate the severity of stuttering and its distribution within age groups figure 3.4 visualizes the uneven distribution of stuttering severity among age groups with older participants displaying more severe stuttering compared to younger participants who show a peak in moderate stuttering. Numbers underline this visualization with the mean SSI-3 value of *M* = 24.00 (moderate stuttering) within young PWS and a mean SSI-3 value of *M* = 29.53 (severe stuttering) for older participants. The comparison of stuttering severity between age groups, however, did not reach significance.

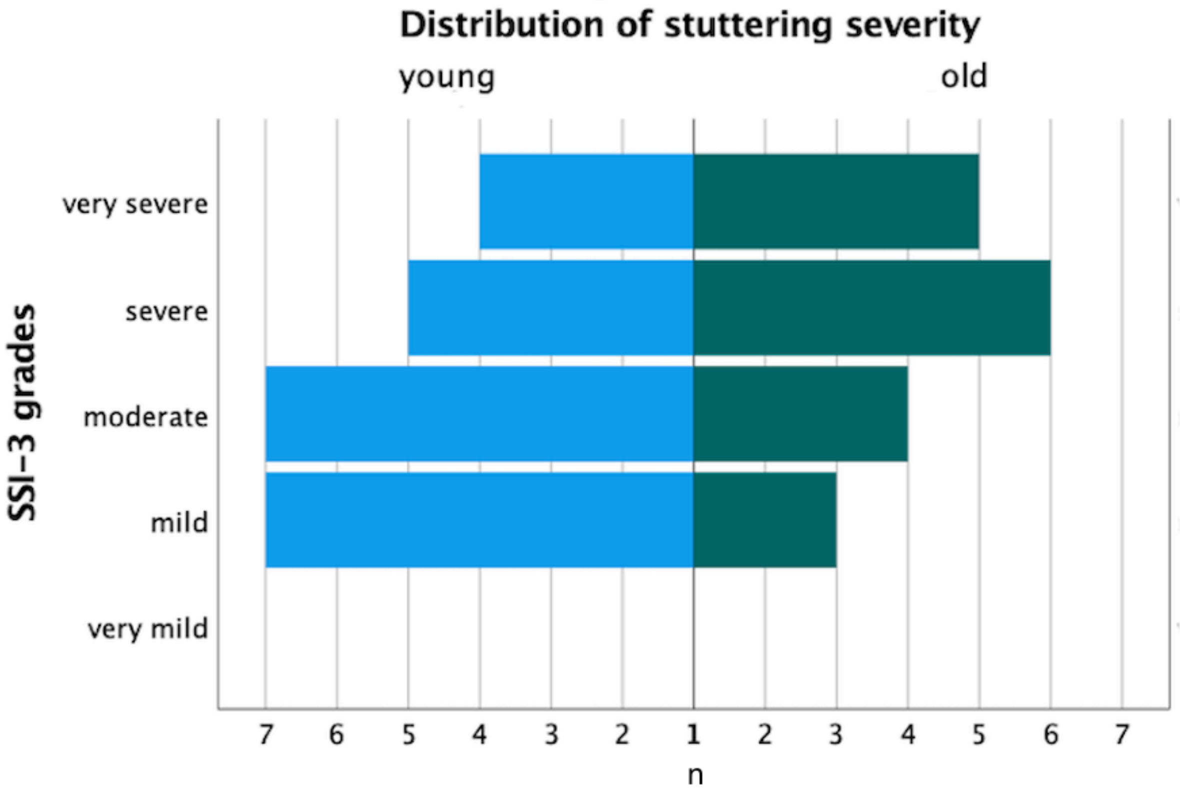


Figure 3.4 Distribution of stuttering severity among young and old participants

In sum, these results on speech rate show lower rates in PWS than in PWNS, as expected. Figure 3.3 depicts speech rates in PWS and PWNS with subdivision into older and younger participants, displaying not only the facts just explained, but also demonstrating the higher variance in speech rate within PWS compared to PWNS, especially when comparing older participants. Furthermore, figure 3.3 shows that within PWNS older participants show a clear rise in speech rate, whereas in PWS, older participants barely show higher rates and – in contrast – even show some very low rates, which might be caused by more severely stuttering participants within the group of older PWS, as figure 3.4 demonstrates.

With regard to the analysis of articulation rate in spontaneous speaking, it is expected to be comparable between groups, since now all instances of disfluencies of any kind (stuttering and other) have been removed as well as pauses linked to disfluencies and pauses > 250ms. However, results of the two-way ANOVA show again a significantly higher rate in PWNS (*M* = 4.38 syllables per second, *SD* = 0.72) compared to PWS (*M* = 3.51 syllables per second, *SD* = 0.96) with *F(*1, 85) = 25.627 and *p* < .001, η^2 = .240. Also between age groups a significantly higher rate in older participants (*M* = 4.19 syllables per second, *SD* = 1.14) was found *F*(1, 85) = 5.759, *p* = .019, η^2 = .066) compared to younger participants (*M* = 3.76 syllables per second, *SD* = 0.72). Again, it is evident that PWS display a higher variability compared to PWNS, as can be seen in Figure 3.5. Despite the removal of all kinds of disfluencies, PWS still show significantly slower rates than PWNS, which hints towards differences within systems of speech motor planning and execution. No interaction was found for articulation rate in speaking.

To further investigate this issue, t-tests were conducted comparing young PWS and PWNS on articulation rate in speaking, displaying significant results with *t*(45) = -3.406, *p* < .001 with slower rates (*M* = 3.43 syllables per second, *SD* = 0.68) in young PWS compared to young PWNS (*M* = 4.08 syllables per second, *SD* = 0.61) as well as old PWS versus old PWNS with *t*(27.662) = - 3.619, *p* < .001 and slower rates but higher variability in old PWS (*M* = 3.60 syllables per second, *SD* = 1.23) compared to old PWNS (*M* = 4.76 syllables per second, *SD* = 0.66). Comparing young and old PWNS with each other, results also show significant differences with *t*(41) = - 3.503, *p* < .001 and slower rates in young PWNS (*M* = 4.08 syllables per second, *SD* = 0.61) compared to old PWNS (*M* = 4.76 syllables per second, *SD* = 0.66). Looking at young and old PWS, though, results are in line with the visualization of data in figure 3.5 with no significant results of the t-test, but a huge variability in rates of old PWS (*M* = 3.60 syllables per second and *SD* = 1.23) compared to young PWS displaying a mean of *M* = 3.43 syllables per second with *SD* = 0.68. The effect of rising rates with age could therefore not be found within PWS – other than in PWNS, where a clear advantage of older participants was evident for the speaking condition. Possible reasons for this phenomenon shall be part of the discussion to follow.

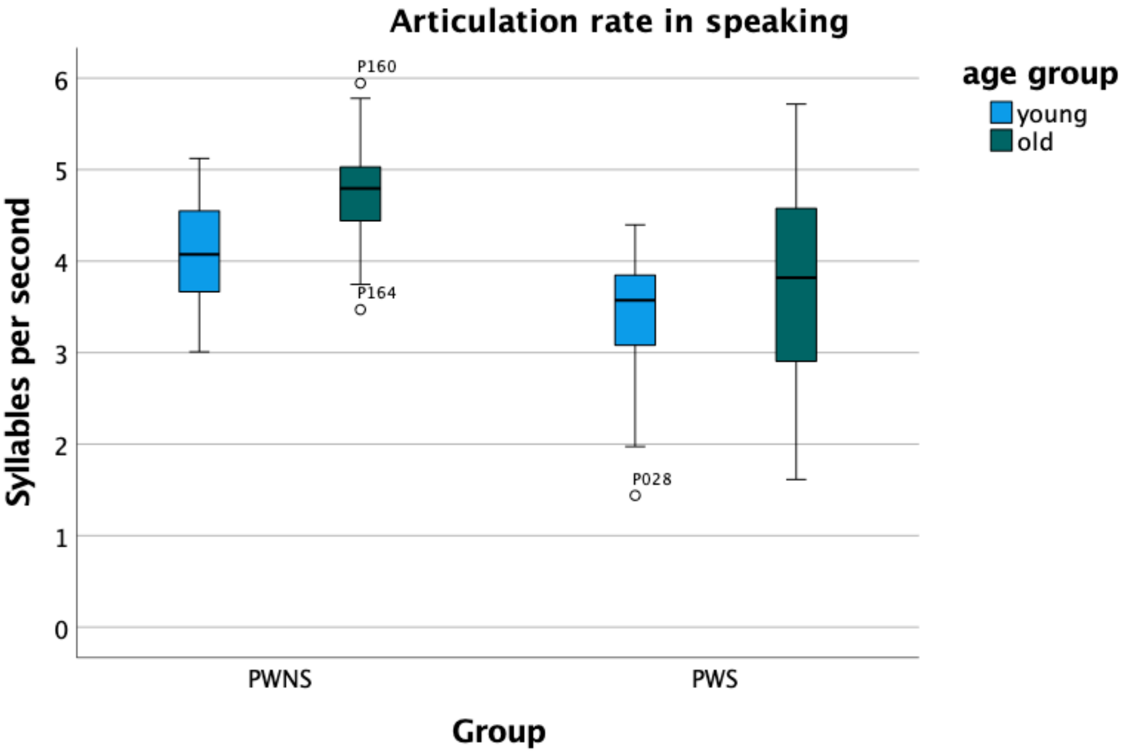


Figure 3.5 Articulation rate in PWNS and PWS in spontaneous speech (interview task). Bars represent speech rate in both groups subdivided for age groups from the first to the third quartile via the median. Whiskers represent all datapoints lying within 1.5 times the interquartile range beyond the quartiles, datapoints above or below that range are marked as outliers.

The influence of age on speech rate and articulation rate are visualized in the two following scatter plots (Figure 3.6a and b). The correlation of age and rate (over both groups) is stronger in articulation rate ($r_s = .357$, $p < .001$) than in speech rate ($r_s = .278$, $p = .010$), probably due to a removal of all pauses and disfluencies. However, looking at correlations for PWS and PWNS separately is far more interesting. Numbers mirror what graphs already display: For both speech rate and articulation rate, the correlations of age and rates are stronger within PWNS (speech rate: $r_s = .632$, $p < .001$; articulation rate: $r_s = .576$, $p < .001$) than in PWS (speech rate: $r_s = .040$, $p = .799$; articulation rate: $r_s = .140$, $p = .375$), probably due to the higher amount of variability, especially shown within older PWS, as discussed before. The effect of significant correlations of age and rates over both groups therefore was mostly carried by PWNS, as the separate correlations have demonstrated. The rise of rates with age and more mature skills in articulation might not be as prominent as expected due to rather low rates caused by symptoms within PWS.

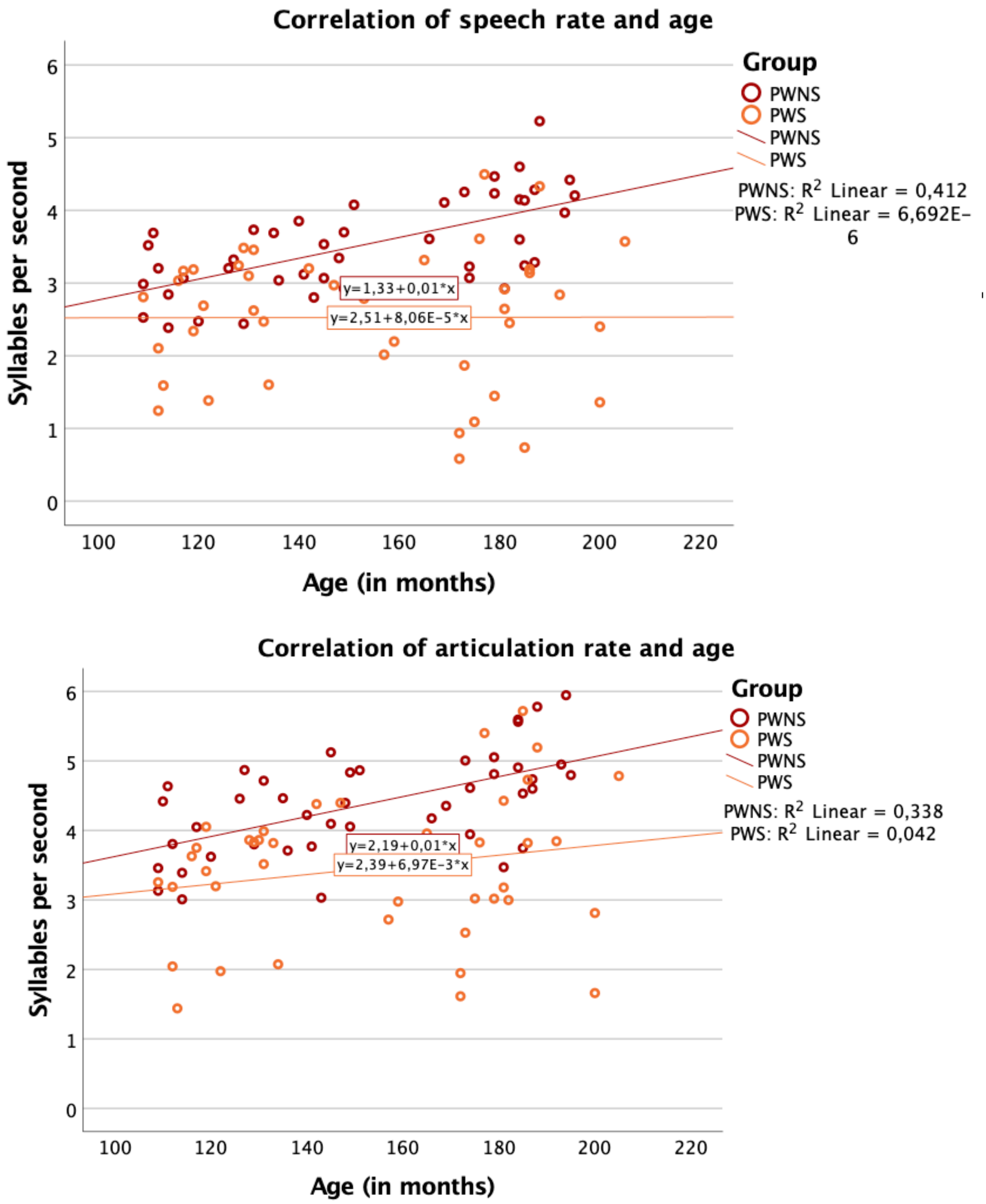


Figure 3.6a and b Correlations of speech rate and articulation rate with age in spontaneous speech (interview task)

Investigating the influence of stuttering severity, as measured by the SSI-3, on speech rate and articulation rate, it becomes clear that negative correlations are very strong, which is obvious for speech rate (r_s = -.655, $p < .001$), since disfluencies are not removed here (see figures 3.7a and b). However, it remains quite strong for articulation rate (r_s = -.416, p = .006) as well, although all instances of disfluencies and pauses related to it had been removed. Looking at correlations for age groups within PWS separately, it becomes evident (as figures 3.7a and b illustrate) that within young PWS this correlation is weaker for speech rate (speech rate: r_s = -.646, $p < .001$) compared to older PWS (speech rate: r_s = -.747, $p < .001$) but stronger for articulation rate ($r_s = -.510$, $p = .013$) compared to older PWS ($r_s = -.464$, $p = .045$). Again, this observation can be explained with the great variability within older PWS, as within this subgroup participants either show high rates due to minor symptoms and better articulatory skills or very low rates with severe symptoms – in the latter case better articulatory skills cannot compensate for the severity of symptoms. Within younger PWS, this spectrum is smaller due to lower rates that come with younger age and less severe symptoms. Concerning articulation rates, where disfluencies of any kind were removed, this variety does not display such a strong impact leading to a rather comparable correlation of young and old PWS. This fact will be discussed in the section following results.

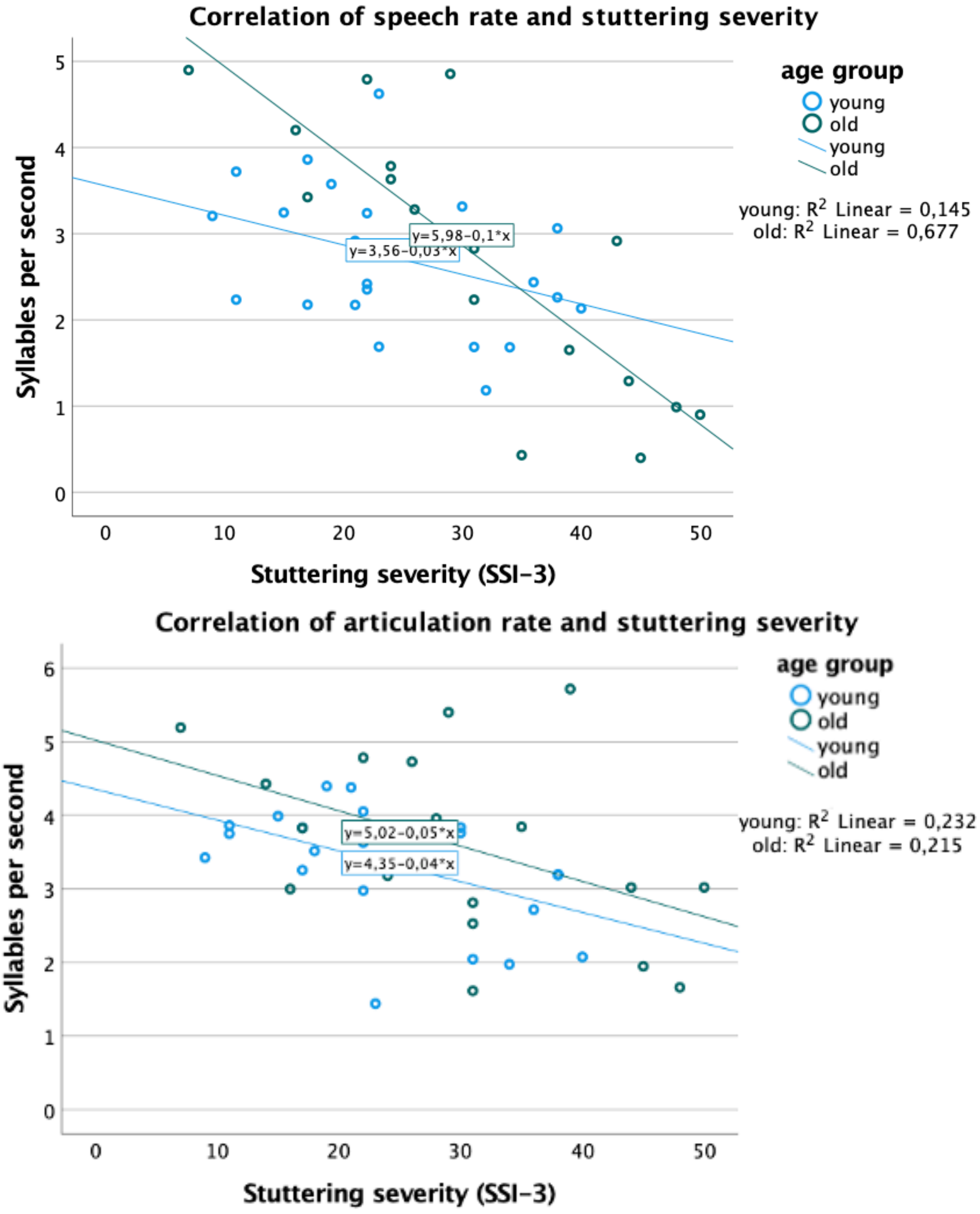


Figure 3.7a and b Correlations of speech rate and articulation rate with stuttering severity as measured with SSI-3 in the speaking task

3.3.2 Results on speech rate and articulation rate in reading

In addition to the exclusion of participants with comorbidities like ADHD or cluttering, participants with dyslexia had to be excluded to avoid falsified results for the reading task. Therefore, participants P22, P24, P52 and P72 were excluded from the analyses.

The first part of this paragraph reports results on comparisons between groups (PWS and PWNS, young and old) on rates during reading of a short excerpt. The second part focuses on possible causes for the differences found between groups. All statistical analyses were, again, conducted in SPSS (Version 27). Normal distribution of the data was controlled for using the Shaprio-Wilk test, showing a normal distribution on speech and articulation rate ($p > .05$) between groups but lacking normal distribution for older participants as the analysis of age groups showed with $p < .001$ for speech rate and $p = .003$ for articulation rate, which is probably due to smaller numbers of participants within this group. The homogeneity of variances was checked, using Levene's test prior to further analyses, which reached significance, indicating no homogenous distribution of variances for either speech rate or articulation rate between groups or age groups. After group comparisons, Spearman-rho correlations were performed to get a better understanding of factors influencing rates.

Results of the ANOVA show a significantly higher speech rate in PWNS than in PWS with $F(1, 78) = 24.012$, $p < .001$, $\eta^2 = .235$ and a mean speech rate of $M = 2.71$ syllables per second ($SD = 1.13$) in PWS compared to PWNS with $M = 3.65$ syllables per second ($SD = 0.76$). Data on speech rate in the reading task displayed significant results ($p < .001$) for Levene's test. Comparing age groups on speech rates, results of the ANOVA also show significance ($F(1, 78) = 5.954$, $p = .017$, $\eta^2 = .071$) with younger participants displaying a slower rate with $M = 2.97$ syllables per second ($SD = 0.71$) compared to older participants ($M = 3.496$ syllables per second, $SD = 1.33$).

Furthermore, the ANOVA showed a significant interaction of group and age group ($F(1, 78) = 4.830$, $p = .031$, $\eta^2 = .058$): Resolving this interaction with the help of figure 3.8, it becomes clear that this interaction is caused by the huge variability shown within the older group of PWS. Whereas within PWNS, older participants show a higher rate compared to younger participants, probably due to more mature reading skills and a more mature articulatory system, this advantage of older participants is not visible at all within PWS: They show a huge variability compared to PWNS as well as compared to younger PWS. This phenomenon is probably caused by more severe symptoms, as it was demonstrated above for the spontaneous speech task and illustrated in figure 3.4. t-tests were conducted to compare those subgroups with each other: Young PWS and PWNS differed significantly on speech rate in reading with *t*(44) = -2.739, *p* = .009 (mean rate in young PWS: *M* = 2.69 syllables per second, *SD* = 0.665, mean rate in young PWNS: *M* = 3.23 syllables per second, *SD* = 0.67) as well as old PWS versus old PWNS with *t*(18.783) = - 3.412, *p* = .003 and slower rates but higher variability in old PWS (*M* = 2.74 syllables per second, *SD* = 1.56) compared to old PWNS (*M* = 4.09 syllables per second, *SD* = 0.46). Comparing young and old PWNS with each other, results also show significant differences with *t*(37.101) = - 3.845, *p* < .001 and slower rates young PWNS (*M* = 3.32 syllables per second, *SD* = 0.90) compared to old PWNS (*M* = 5.15 syllables per second, *SD* = 0.49). Looking at young and old PWS, though, results of the interaction found in the ANOVA and visualized in figure 3.8, are replicated with no significant results of the t-test but a huge variability in rates of old PWS (*M* = 2.74 syllables per second and *SD* = 1.56) compared young PWS displaying a mean of *M* = 2.69 syllables per second with *SD* = 0.66. The effect of rising rates with age could therefore not be found within PWS – other than in PWNS, where a clear advantage of older participants was evident for the reading condition.

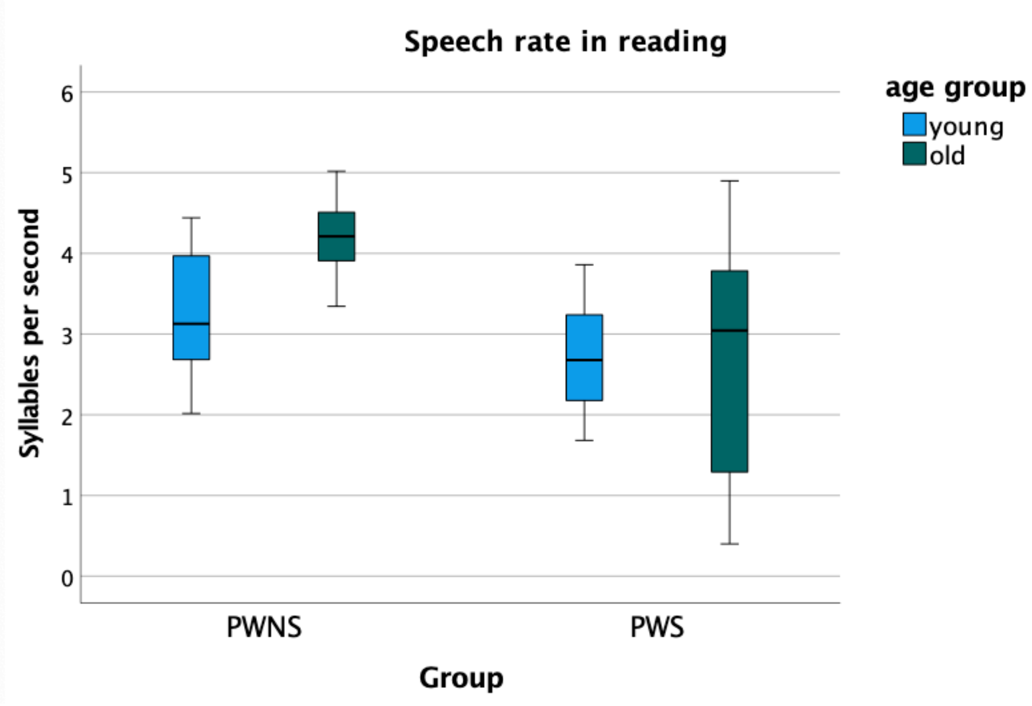


Figure 3.8 Speech rate in PWNS and PWS in the reading task.
Bars represent speech rate in both groups subdivided for age groups and display the median; details as in figure 3.3.

Results on articulation rate in the reading task lacked homogeneity of variances as well. Comparing PWS and PWNS, differences between groups are highly significant *F*(1, 78) = 27.540, *p* < .001, η^2 = .261 with PWS showing significantly lower rates with *M* = 3.70 syllables per second (*SD* = 0.99) compared with *M* = 4.691 syllables per second (*SD* = 0.855) in PWNS.

Comparing age groups, young participants displayed a lower mean rate with *M* = 4.01 syllables per second (*SD* = .092) than older participants (*M* = 4.48 syllables per second, *SD* = 1.14), as significant ANOVA results implied with *F*(1, 78) = 4.996, *p* = .028, η² = .060.

An interaction for age group and group was found here as well, however, it was only marginally significant with *F*(1, 78) = 4.071, *p* = .047, η² = .050. This weak interaction can be resolved with the help of figure 3.9, showing a high variability within older PWS even though it appears not as high as in speech rate (see figure 3.5). Still, it is evident that in PWNS older participants display higher rates compared to younger and this effect cannot be reported for PWS, where older participants display the highest variability of all groups – despite the removal of all kinds of disfluencies.

In sum, the comparison of speech and articulation rates in reading has again demonstrated, that within PWS, older participants differ significantly in their performance not only from age matched PWNS, but also from younger PWS. Comparing those subgroups separately, this fact is supported by significantly differing rates between young PWS and PWNS on articulation rate with *t*(44) = -2.471, *p* = .017 (mean rate in young PWS: *M* = 3.68, *SD* = 0.83, mean rate in young PWNS: *M* = 4.32 syllables per second, *SD* = 0.90) as well as old PWS versus old PWNS with *t*(20.564) = - 4.328, *p* < .001 and slower rates but higher variability in old PWS (*M* = 3.73 syllables per second, *SD* = 1.19) compared to old PWNS (*M* = 5.07 syllables per second, *SD* = 0.45). Comparing young and old PWNS with each other, results also show significant differences with *t*(37.101) = - 3.845, *p* < .001 and slower rates young PWNS (*M* = 4.32 syllables per second, *SD* = 0.90) compared to old PWNS (*M* = 5.15 syllables per second, *SD* = 0.49. Analyzing data of young and old PWS, though, results of the interaction found in the ANOVA and visualized in figure 3.9 are confirmed with no significant results of the t-test but a huge variability in rates of old PWS (*M* = 3.73, *SD* = 1.19) compared to young PWS displaying a mean of *M* = 2.68 syllables per second with *SD* = 0.83. The effect of rising rates with age could therefore not be found within PWS – other than in PWNS, where a clear advantage of older participants was evident for the reading task.

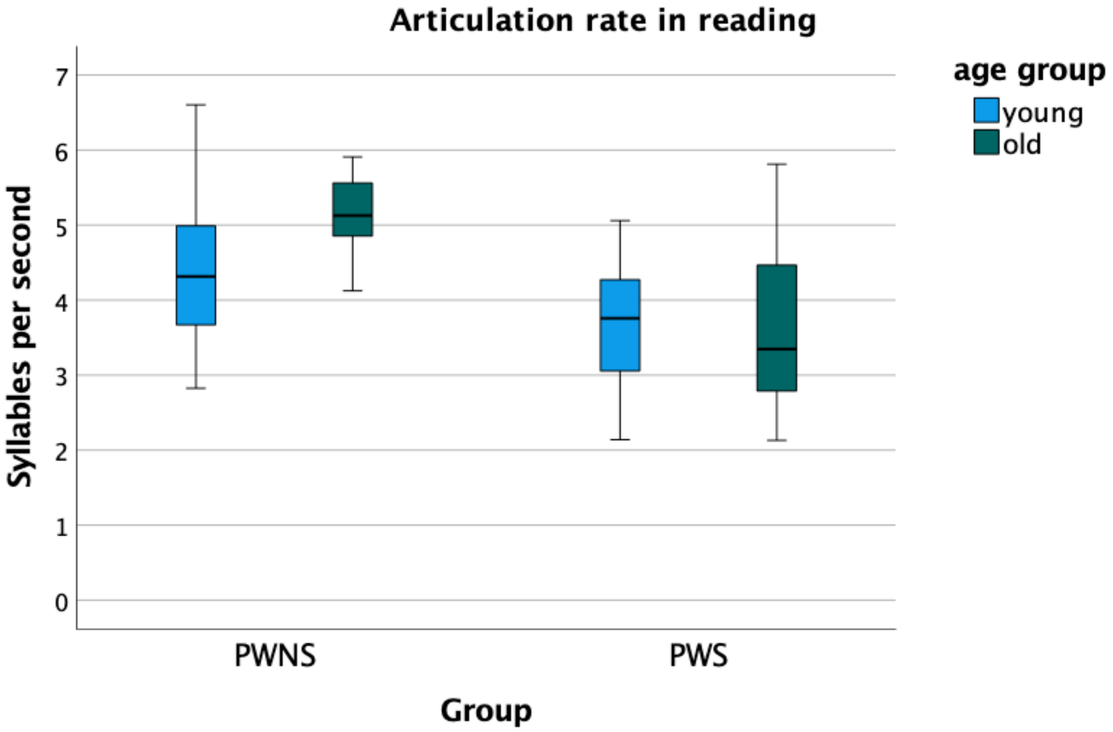


Figure 3.9 Articulation rate in PWNS and PWS in the reading task.
Bars represent speech rate in both groups subdivided for age groups and display the median; details as in figure 3.3.

The influence of age on this task is even more interesting, compared to rates in speaking since reading is a competence that develops with age, as explained before. Therefore, correlations of age and rates in the reading task are supposed to indicate how strong the factor age influenced performance on this task. For speech rate the correlation with age was quite strong already ($r_s = .433$, $p < .001$), indicating a higher rate with rising age. However, as figure 3.10a shows, this applies more for PWNS than for PWS, where rate does not increase as much due to various (older) participants with an extremely low rate despite the removal of disfluent parts. This observation shall be discussed later as part of the question concerned with a compromised speech system in PWS. In articulation rate (figure 3.10b) the correlation is moderate ($r_s = .366$, $p < .001$), also hinting towards a higher rate with rising age. Here as well, this applies more for PWNS than for PWS, where mostly older participants flatten the curve because of lower rates, probably for the same reason as mentioned above. Observing those differences between groups (see figure 3.10a and b), it seems more interesting to look at correlations for each group separately, instead of using a correlation with both groups merged: Numbers display what graphs already demonstrate: For both speech rate and articulation rate, the correlation of age and rates is stronger within PWNS (speech rate: $r_s = .741$, $p < .001$; articulation rate: $r_s = .671$, $p < .001$) than in PWS (speech rate: $r_s = .146$, $p = .375$; articulation rate: $r_s = .094$, $p = .570$), where no significant correlation was found - probably due to the higher amount of variability, shown within older PWS especially. The effect of significant correlations of age and rates over both groups, therefore, was carried mostly by PWNS and not PWS, as the separate correlations have demonstrated. The rise of rates with age and more mature skills in articulation and reading are layered by rather low rates caused by symptoms within PWS influencing articulation rate as well, despite the removal of all kinds of disfluencies.

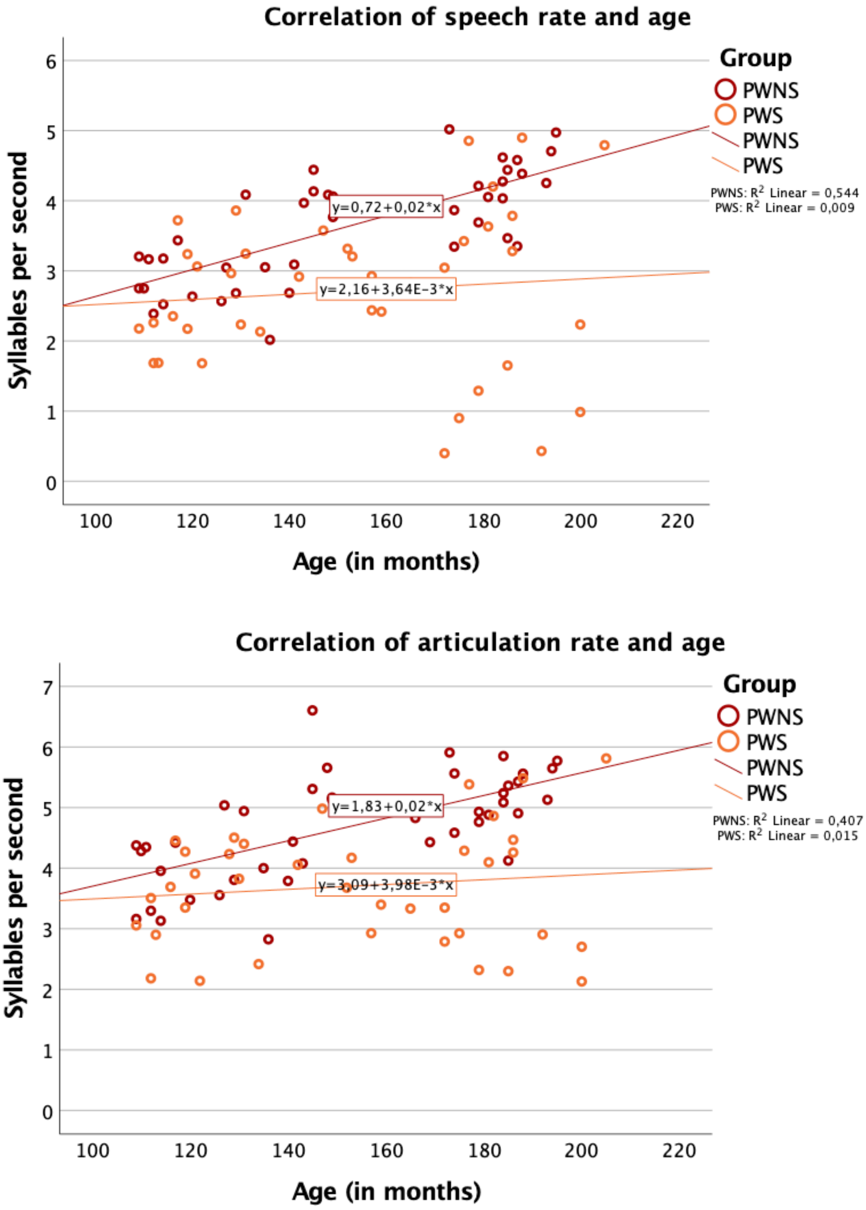


Figure 3.10a and b Correlation of speech rate and articulation rate with age in the reading task

To investigate the relation of stuttering severity and its influence on speech or articulation rate more precisely, correlations of stuttering severity and rates were calculated, indicating a very strong correlation in both cases: In speech rate the correlation with stuttering severity as diagnosed with the SSI-3 was very strong (r_s = -.628, p < .001) and comparable to the correlation of articulation rate and SSI-3 with r_s = -.665, p < .001. Results, therefore, demonstrate the strong connection between initially diagnosed stuttering and rates within the reading task, with rates declining as stuttering severity rises. Analyzing correlations for age groups within PWS separately, it becomes evident (as figures 3.11a and b illustrate) that within young PWS this correlation is weaker for speech rate (speech rate: r_s = -.414, p = .056) and articulation rate (r_s = -.503, p = .017) compared to older PWS for speech rate (r_s = -.867, p < .001) and articulation rate (r_s = -.872, p = <.001). This observation can be explained with older PWS having more severe symptoms. The mere factor age that leads to more mature articulatory and reading skills does not have any impact here, since the severity of symptom overlays the advantage of age. Even in the case of the reading task, where older participants were expected to show higher rates compared to younger, this is not true for the group of PWS. This fact will be discussed in the section following results.

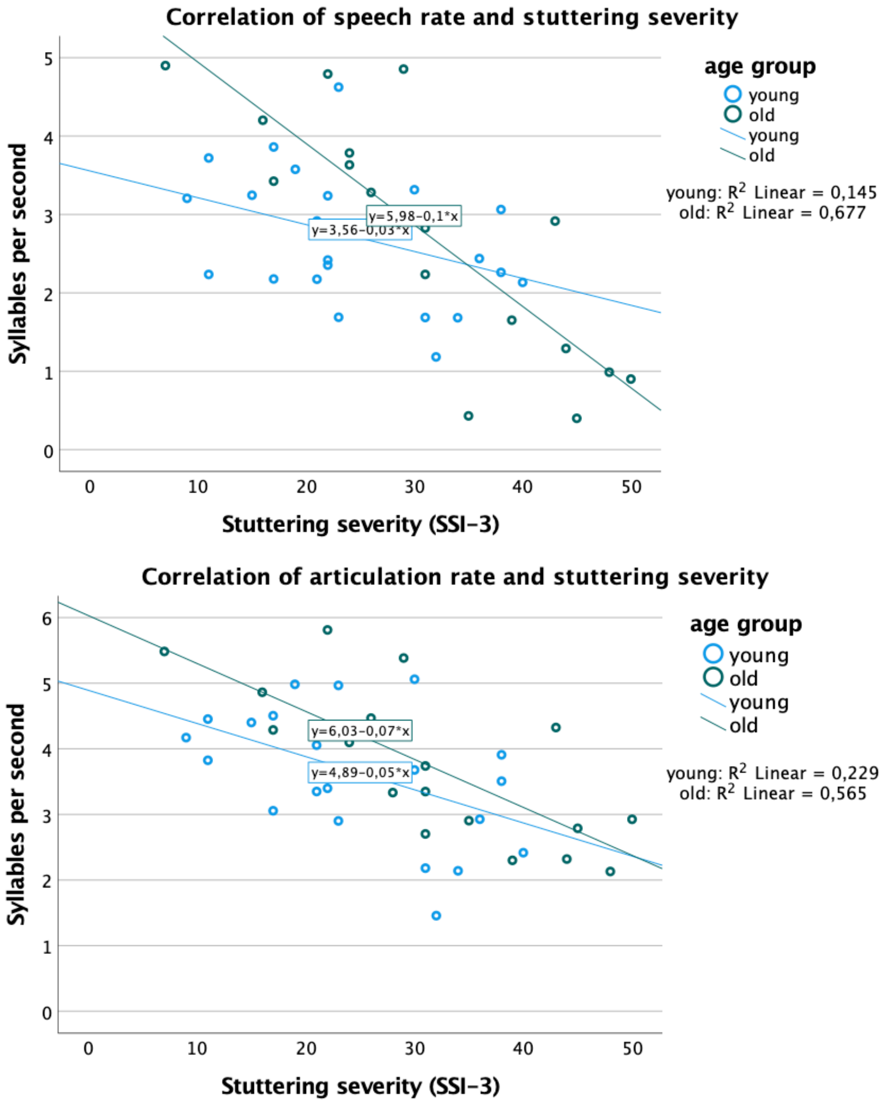


Figure 3.11a and b Correlation of speech rate and articulation rate with stuttering severity (as measured with SSI-3) in the reading task

Having completed the analyzes of speech rate and articulation rate in both tasks, speaking and reading, the two modalities will now be compared to each other directly.

3.3.3 Comparison of rates in speaking and reading

After a comprising analyse of the two modalities separately, correlations with each other shall be conducted to investigate the relation of speaking and reading with each other as well as the influence on each other. It is hypothesized that higher rates in speaking lead to higher rates in reading as well. However, for younger participants this relation must be investigated separately to account for possible effects of reading fluency.

In analogy to chapter 3.3.2, participants P22, P24, P52 and P72 were excluded from the analyses due to dyslexia which could have falsified results regarding rates in reading tasks that are also being compared within this paragraph.

As figures 3.12a and b illustrate, there is a strong correlation of speech rates in speaking and reading ($r_s = .726$, $p < .001$) and an even stronger correlation of articulation rates in speaking and reading ($r_s = .763$, $p < .001$) indicating that there is a strong link between rates in the different tasks. As figures 3.12a and b also demonstrate, rates are higher in PWNS than in PWS, as discussed above. When conducting the same correlations only within the group of PWNS compared to PWS, it becomes evident that results are comparable to overall group results and stuttering has only a minor influence on the correlation of rates: Whereas in PWNS(speech rate $r_s = .569$, $p < .001$; articulation rate $r_s = .728$, $p < .001$) speech rate shows a slightly smaller correlation than within the complete group, in PWS (speech rate: $r_s = .730$, $p < .001$; articulation rate $r_s = .655$, $p < .001$) articulation rates display a weaker correlation. These results imply that stuttering only has a negligible impact on the correlation of rates in the two given modalities and that for both groups, PWS and PWNS it can be stated that a higher rate in speaking is significantly correlated with a higher rate in reading – and vice versa.

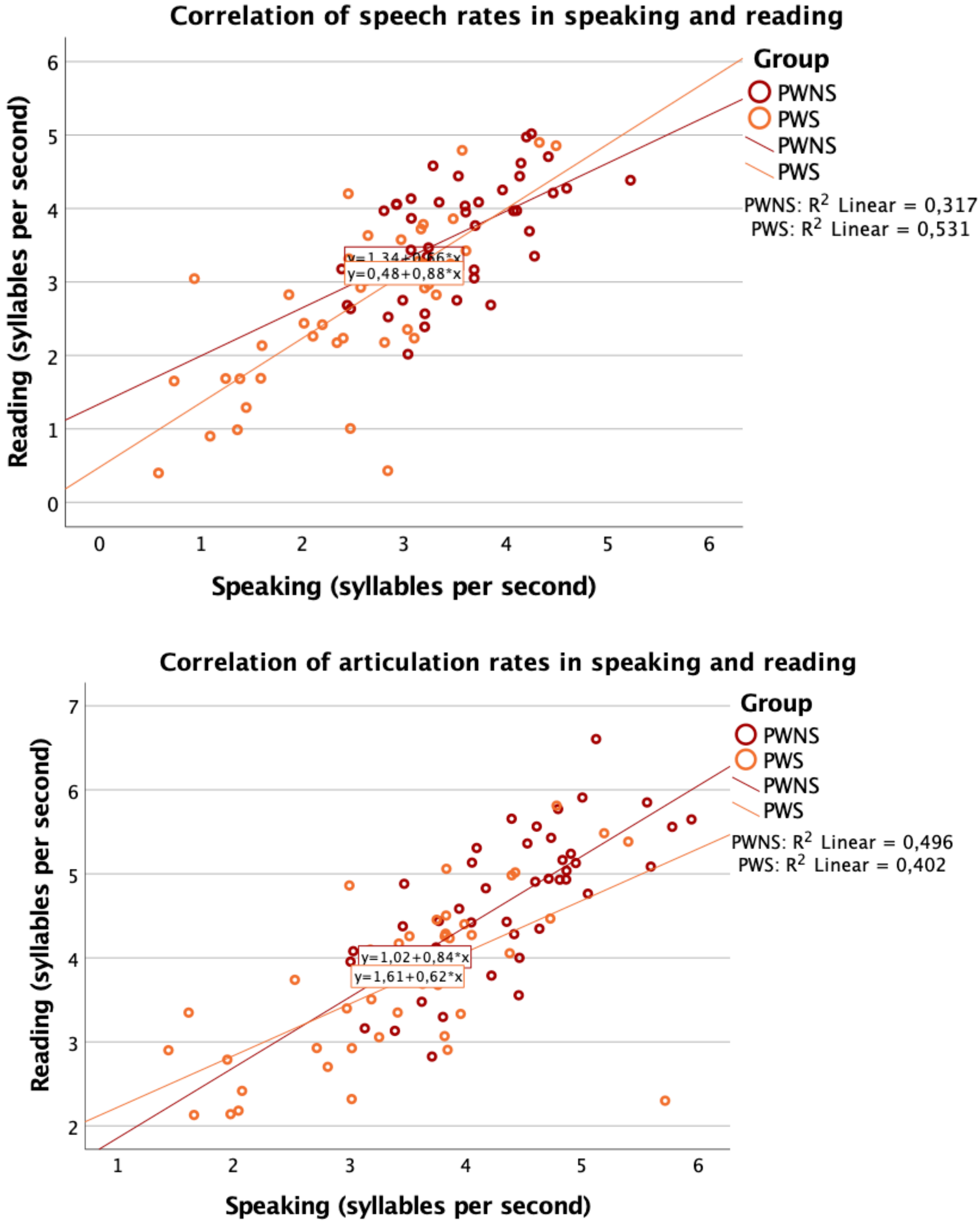


Figure 3.12a and b Correlations of rates in reading and speaking

Regarding the influence of age on the correlation calculated above, analyzes on young and old participants separately came to the following results: Young Participants displayed significant correlations for speech rates ($r_s = .576$, $p < .001$) and articulation rates ($r_s = .743$, $p < .001$). For old participants the same analyzes were conducted: the correlation of speech rates in speaking and reading is higher in old participants ($r_s = .749$, $p < .001$) indicating that in old participants a higher rate in speaking was associated with a higher rate in reading, whereas in articulation rate ($r_s = .691$, $p < .001$) it was the other way round with older participants displaying a weaker correlation than younger participants. This might hint towards the fact that the two modalities (speaking and reading) are not as closely linked in older participants, maybe due to the previously observed variability, that was still observable in articulation rate, where all disfluencies had been removed.

In sum one can say that independently of group or age group, correlations of speech rates in speaking and reading as well as correlations of articulation rates in speaking and reading are quite high. These results suggest that high rates in in speaking are associated with high rates in reading and vice versa.

Having completed the analyzes of speech rate and articulation rate as well as a comparison of both modalities, the results will now be discussed and related to existing studies in the following section.

3.4 Discussion

The aim of this study on speech and articulation rate in PWS and a fluently speaking control group was to explore whether PWS differ from PWNS. Comparisons between those two groups focused on speech rate, as a more global measure of verbal output and on articulation rate, a rather specific term to quantify motor planning and execution time to gain information about possible differences between those two constructs that may allow to draw conclusions about similarities and differences between the groups tested and their mechanisms of speech production. A further aim was to compare age groups: children (“young”) and adolescents (“old”) were investigated to gain insight into differences that might be caused by age and the development of speech systems as well as the development of stuttering with age. On top of that, analyses were based on participants’ spontaneous speech during the natural setting of a conversation and on their reading performance when reading a popular German children’s book aloud. Hence, in addition to the comparison of groups and age groups, two different tasks were compared here.

Comparing groups regarding general parameters such as their mean age, inhibitory control or working memory it is clear that PWNS and PWS show comparable results. Age groups are with $n_{\text{young}} = 48$ and $n_{\text{old}} = 38$ not perfectly comparable, the distribution within PWS and PWNS each is $n_{\text{young}} = 24$ and $n_{\text{old}} = 19$. This factor must be kept in mind when discussing results on age group comparisons.

When investigating speech and articulation rate in speaking within PWS, various results seem possible as previous studies on this topic have demonstrated: Whereas some authors could not find differences in rates between PWS and PWNS, including school-aged children (Franke et al., 2021), others found faster rates in children later diagnosed with stuttering compared to fluently speaking children (Kloth et al., 1995), or the exact opposite, slower rates (Logan et al., 2011). For the interview task where participants were speaking in a natural setting, speech rate analyses displayed significant differences with PWS showing lower rates compared to PWNS. This result is not surprising, since, in speech rate, dysfluencies of any kind that interrupt the flow of speech are not removed and results are in line with previous studies on this topic (Bloodstein, 1944; Logan et al., 2011; Meyers & Freeman, 1985). The authors found that the severity of stuttering is linked directly to decreases in speech rate (Meyers & Freeman, 1985), as was demonstrated for PWS in this study as well. Speech rate is a relevant parameter when diagnosing stuttering or quantifying therapeutic success, since a strong deviation from speech rate of PWNS is a clear sign that verbal output is affected and at the same time an approximation in speech rate to rates of PWNS indicates a normalization of verbal output. However, this fact is not surprising, since the causality is quite clear: With more severe stuttering symptoms there are two ways of dealing with it. One is to slow down the rate as a compensatory strategy to avoid symptoms, the other is to simply show interruptions and both lead to the same result: a lower verbal output as measured in syllables per seconds.

The comparison of age groups showed only a marginal significance with the ANOVA which therefore is rather to be interpreted as a tendency for older participants to have higher rates. This result might seem surprising at first sight, since rate is expected to rise with age, as articulatory systems get more mature. In the case of the participants in this study, this effect might reflect an interaction with the fact that older participants also seem to suffer from more severe stuttering symptoms with a mean SSI-3 value of $M = 24.00$ (moderate stuttering) within young PWS and a mean SSI-3 value of $M = 29.53$ (severe stuttering) for old participants. The comparison of stuttering severity between age groups, however, did not reach significance and correlations on stuttering severity and age also failed to reach significance. On top of that, the effect of age might be more strongly visible for the task of reading, since reading skills are increasing faster than skills in speaking, which are developed earlier (A. H. Chen et al., 2019).

The higher variability in speech rates in PWS can be explained with stuttering symptoms: Whereas minor symptoms only have a minor impact on rate, participants with severe symptoms display a strongly affected rate, leading to a broader spectrum of rate variability within PWS.

For the analysis of articulation rate in spontaneous speaking, one might not expect significant differences between PWS and PWNS, since in articulation rate, there should be no bias caused by stuttering symptoms. Results on articulation rate in stuttering have been inconclusive; a very recent study with comparable methods as to the study at hand, reported comparable results between PWS and PWNS (Franke et al., 2021). However, Franke and colleagues used verbal output from a reading task only and not a combination of reading and speaking. In the thesis at hand, however, results on articulation rate show significant differences between groups, with PWNS displaying higher rates. Hence, stuttering severity showed direct impact on perceptually fluent articulation rate in PWS, leading to a lower rate compared to PWNS – a finding that was supported by correlations of stuttering severity and articulation rate. As in speech rate, a higher variability, especially in adolescent PWS is evident within articulation rate, demonstrating the strong influence of stuttering on perceptually fluent parts of speech, that might seem unimpaired at first sight. Exactly this influence of stuttering on perceptually fluent or symptom-free speech was demonstrated before in preschoolers (Tumanova et al., 2011) The phenomenon of a higher variability in articulation rate was also demonstrated before (Kloth et al., 1999). Both explanations seem reasonable here: The lower rate in perceptually fluent and, therefore, supposedly unimpaired speech could either derive from additional time in processing linguistic and phonological information to plan speech movement or from certain trained and sometimes subconscious compensatory strategies to avoid stuttering symptoms, such as a slower fluent articulation. So far, it cannot be clarified which reason seems more plausible, but it is beyond all question that results imply a compromised speech system in PWS. As in speech rate, the comparison of articulation rate in speaking between age groups showed a significantly higher rate within older participants as calculated with the ANOVA. As t-tests have shown, the difference between age groups was significant for PWNS but not for PWS, indicating that this variability leads to smaller or even insignificant effects when comparing age groups.

Generally, the factor age had a strong impact on both, speech and articulation rate as significant correlations had demonstrated above. However, this fact seems to be truer for PWNS than for PWS as figure 3.6a and b imply. Whereas in PWNS the relation of rates and age develops linearly, showing significant correlations, the relation of rates and age is not linear within PWS, as correlations confirm (no significant correlation within PWS). Age was not so much expected to have a strong influence on rates in speaking, but rather on rates in reading, since reading is a skill that is still being developed within the age groups investigated (Landerl & Wimmer, 2008). But as mentioned before, it seems that stuttering severity had strong impact on the relation of rates and age and therefore, the correlations of stuttering severity and rates needs further discussion: a strong negative correlation for speech rate seems reasonable, since interruptions and pauses caused by stuttering symptoms decrease the rate. The more severe symptoms are, the smaller speech rate naturally gets. After the removal of all kinds of disfluencies – stuttering and other – it seems like stuttering severity should not have a strong impact on rate anymore.

Correlations on articulation rate and stuttering severity demonstrate the opposite: These findings suggest that even the perceptually fluent speech of PWS is influenced by their stuttering – although stuttering might seem absent at the moment of investigation. Still, psycholinguistic models claiming that PWS show slower rates due to longer processing of linguistic and phonological information as well as extra time needed for the planning of upcoming speech movements, seem reasonable when explaining the results above. Furthermore, it seems plausible that not only extra time in processing and planning is required, but also strategies to avoid symptoms might have developed such as rephrasing of sentences, exchanging of certain words and maybe just a slower rate in general.

Results on the reading task add interesting information on the topic investigated, since reading aloud is a competence that adds further demands on various language capacities. It can be hypothesized that immature or challenged speech (motor) systems display more difficulties in reading; in the present study this would apply to younger PWS. Results on speech rate during reading showed a significantly lower rate in PWS compared to PWNS as well as a significantly lower rate within young versus old participants. Those results go along with the hypotheses and support the idea of compromised speech systems (stuttering and/or immaturity) displaying strongest effects on reading rate. Since reading fluency not only requires fluent articulation, but also successful text decoding, it seems reasonable that rates of populations affected by those constructs – PWS and young speakers – reflect this fact. Within the reading task, as well, a greater variability in older PWS is clearly visible. The advantage of age in this task, as figure 3.10a and b shows within PWNS, is barely visible for older PWS which is probably caused by interruptions such as repetitions, prolongations, and blocks, but also by a slower rate as a compensatory strategy. After the removal of all instances of disfluencies and pauses directly linked to it as well as all pauses > 250ms, groups of PWS and PWNS still differ significantly on mean articulation rate with PWNS articulating more syllables than PWS. Between age groups, differences on articulation rate reached significance as well, although, in case of age group comparisons significance was a lot weaker. In sum, the expectation of lower rates in young participants due to challenges of reading aloud can be claimed as fulfilled and is in line with comparable studies (Franke et al., 2021). Again, the results of PWS showing lower rates despite the removal of all kinds of disfluencies in addition to the big variability, especially within older PWS, leaves the question of reasons for this phenomenon. It seems that with growing severity of symptoms the mere exclusion of these is not enough to achieve comparable rates between PWS and PWNS probably due to compensatory strategies or different systems of speech planning and production, as mentioned before.

The influence of age on this task is very interesting, since it might be stronger than in the interview task, as reading is a skill that improves within the age span tested. Indeed, results showed strong correlations of age and speech and articulation rate; however, they did not exceed correlations for the interview task. Comparable to analyses of the interview task is the fact that the correlation seems to be carried by PWNS, since the relation of rates and age is clearly stronger and more linear in PWNS than in PWS. Within the group of PWS, older participants with low values in rate seem to be flattening this linearity. Although there was no significant correlation of age and stuttering severity it still seems to bias the picture as some of the most severely affected PWS are older participants. In sum, as lines within figures 3.10a and b indicate, age strongly correlates with speech and articulation rate, although this fact applies more to PWNS than to PWS. Therefore, calculations on the relation between stuttering severity, as measured with SSI-3 and rates were conducted, displaying strong negative correlations, with speech rate as well as with articulation rate in reading. In sum, the fact that even articulation rate is strongly influenced by stuttering severity seems like a common thread indicating different systems of speech planning and execution in stuttering – independent from the task that generated verbal output.

Disfluencies are naturally found in every person’s speech without thinking of it as something pathological. Therefore, also the fluently speaking participant’s speech was processed and disfluencies and pauses were excluded for analyzing articulation rate. In stuttering, however, the rate of disfluencies is abnormally high leading to the classification as a speech disorder (Arbisi-Kelm & Jun, 2005). Hence, in most cases the distinction between the speech of PWS and PWNS is quite clear. After the removal of all instances of disfluencies, though, one would expect that there is no difference to be found between the speech of PWS and PWNS. Results, however, indicated the opposite. One study investigating this topic, could not find significant differences in articulation rate between PWS and PWNS, but found pauses in PWNS to be shorter than in their matched peers who stutter. Furthermore, breath pauses were more frequent in PWS (Franke et al., 2021). Pauses smaller than 250ms were not excluded for analyses on articulation rate. It therefore might seem possible that many small “undetected” pauses contribute to the significant difference in articulation rate between PWS and PWNS. However, it is not only in terms of pauses that PWS differ from fluently speaking peers in their perceptually fluent speech: Max and Gracco (2005) found longer durations from the bilabial closing to vocal fold vibration. A cineradiographic study by G. N. Zimmermann (1980) replicated results in terms of longer duration from movement onset to peak velocity and in VOT as well as a longer remaining of lip and jaw within the steady state position when articulating vowels; on top of that Zimmermann also found a greater asynchrony in the organization of lip and jaw movement. Those results of the perceptually fluent speech of adult PWS indicate that many fine-tuned processes within speech production - that are of course not apparent when analyzing speech or articulation rate as was done in this study - may in sum result in measurable effects like a slower articulation rate within PWS. Another study by McClean, Kroll, and Loftus (1990) compared lip and jaw movement within the perceptually fluent speech of PWNS, PWS without a previous therapeutic intervention and PWS who received previous therapeutic intervention. They found an increased jaw movement duration in PWS after therapy and concluded that those anomalies found are the result of adjustment acquired in therapy rather than causal aberration on neurological level. Although participants of the study at hand were explicitly tested prior to therapy the possibility of self-trained compensatory strategies cannot be excluded. Adding up those results, it seems quite reasonable to find significant differences within the perceptually fluent speech of PWS, since aberrations in fluent speech can appear so minimally and at such specific levels that are not recognized and considered when measuring articulation rate.

Adding the potential effect of cognitive effort needed for the tasks presented – answering questions in an interview and reading a text excerpt aloud, results are in line with Erdemir et al. (2018) who found articulation rate to be significantly lower in PWS compared to fluently speaking peers in a task with emotional load (story retelling after watching an emotionally arousing film sequence). As hypothesized before, these results could hint towards a more instable and immature speech-motor system in PWS where (emotionally) complex situations lead to interferences with closely located areas in the brain causing stuttering symptoms due to a disturbed transport of information (Johnson et al., 2010). And as results of this study have shown, stuttering severity of a participant had strong influence on his fluent articulation rate since they spoke at a significantly slower rate.

From a neural point of view, it seems reasonable that aberrant patterns of activation as well as structural deviations in the brain of PWS do not just disappear as one analyzes verbal output after the exclusion of disfluencies (Chang et al., 2008; Chang et al., 2015; Chang & Zhu, 2013). As many studies on the neurological underpinnings of stuttering have shown, it takes intense therapy to lead to a neural reorganization that resembles the neurological organization of PWNS (Lu et al., 2012; Lu et al., 2017). Therefore, it seems reasonable that even the perceptually fluent speech of PWS shows subtle deviations from typically fluent speech, as the origin of speech and its neural correlates are also deviant.

When interpreting and discussing the results, some limitations of the study must be mentioned as well: The chosen text excerpt was well suited for the age span investigated in this study, however, not every participant had read this excerpt prior to therapy which is why for some participants another comparable text was used for the analyzes. This fact might weaken the comparability of stimuli and results for the reading task. When comparing age groups, younger participants outnumber older participants which is not ideal for the comparison of results between age groups. However, the number is perfectly balanced when comparing PWS and PWNS which was more important for the questions raised in this thesis.

In sum, the work in hand presents preliminary results on a topic that still needs further research, since there is a lack of data, especially within the population of young PWS. Results have demonstrated that PWS differ significantly in speech and articulation rate from fluently speaking peers. This was true for spontaneous speaking during the interview task as well as for the reading task. Comparing speech rate between PWS and PWNS, it was not surprising to find significant differences between groups. In articulation rate, however, results hint towards differing systems in speech planning and execution, as significant differences between the perceptually fluent speech of PWS and PWNS indicate. Perceptually fluent speech in PWS obviously is not comparable to PWNS and furthermore it is not yet fully understood. Perceptually fluent speech of PWS is not just the same as the speech of PWNS, it rather seems that – even in the absence of stuttering symptoms – the verbal output of PWS differs in many subtle ways that, depending on the method of investigation, might stay hidden. Identifying the exact qualities of (perceptually fluent) stuttered speech is an immense challenge that must be met in future research. One step into this direction is taken in the chapter to follow.

4. Verbal timing deficits under fluency enhancing conditions in children and adolescents who stutter

Knowing about the differences and aberrations in stuttered speech, we are interested in the potential differences in the speech of PWS that is perceptually fluent. After chapter 3 has demonstrated already that merely the exclusion of symptoms does not lead to a comparable performance on articulation rate between PWS and PWNS, perceptually fluent verbal output will be analyzed in detail. Hypotheses as well as results will be discussed with respect to collected literature on stuttering and rhythm at the behavioral and neurological levels. Therefore, the introduction revisits the most important findings of the previous chapters to prepare the reader for the analyses and results of this study on verbal timing deficits.

4.1 Introduction

Research has made enormous progress in the last decade in understanding the human ability for perceiving, processing, and producing rhythm. However, when it comes to the rhythmic processes active during speech production, there are still many unresolved questions – especially within the subgroup of persons affected by stuttering (Ravignani et al., 2017). One line of research proposes that a general internal timing deficit underlies stuttering since people who stutter show altered patterns of verbal and non-verbal synchronization abilities that can mostly be described as a less accurate, more variable rhythmic performance and an over-anticipation in synchronization (Falk et al., 2015; Olander et al., 2010; Sares et al., 2019). One very interesting finding by Olander and colleagues (2010) is that the clapping of hands and the synchronization of the clapping to a beat showed significantly more variability in CWS than in CWNS. To be more specific, in 60% of CWS the variability was the same as in CWNS, but in 40% the CWS showed a poorer performance in clapping than the worst of CWNS (Olander et al., 2010). Another study compared the synchronizing performance of children and adolescents who stutter to those who do not stutter in a finger tapping task with simple and complex musical beats (Falk et al., 2015). Throughout three different inter-stimulus-rates (450ms, 600ms and 750ms) CWS showed a poorer synchronization performance concerning the accuracy and the variability as compared to CWNS. A further finding was the fact that CWNS improved in their synchronization ability with age, whereas CWS did not. Additionally, there seemed to be a link between a low synchronization consistency and a higher stuttering severity, which led the authors to the assumption that there seems to be a generalized timing-deficit underlying developmental stuttering (Falk et al., 2015). Contradictory results however, by Hilger, Zelaznik and colleagues (2016) show quite the opposite pattern. By investigating 115 CWS and 45 CWNS on a bimanual handclapping task and re-testing the same cohort every year for six years (starting at an age of 3;5 to 9;5 years) the authors not only compared differences between groups, but also examined the potential relationship between rhythmic abilities and stuttering persistency. They could neither replicate the findings of the preceding study by Olander and colleagues (2010), i.e. they did not find differences in the variability of the inter-clap-interval, nor could they demonstrate a relationship between the performance on the motor timing task and stuttering recovery/persistency. Behavioral results in sum seem inconsistent, maybe due to the heterogeneity of stuttering, differences in the age groups investigated, or the task under investigation.

Another line of research has found various aberrations, structural and functional, when it comes to the neural correlates of speech and rhythm in PWS (Chang, Chow, Wieland, & McAuley, 2016; Chang & Zhu, 2013; Fujii & Wan, 2014; Misaghi, Zhang, Gracco, De Nil, & Beal, 2018; Sowman et al., 2014). In the last years of research on the causes and neurological underpinnings of stuttering, a great deal of progress has been made in finding differences between PWS and PWNS: Not only in brain structure, but also in brain function significant differences have been found in auditory and motor regions: During speaking tasks - paced and unpaced (Sowman et al., 2014; Toyomura et al., 2011) - in rest (Chang et al., 2015; Xuan et al., 2012) and also regarding the connectivity within and between the areas relevant for auditory and motor processing (Cai et al., 2014; Chang & Zhu, 2013; Misaghi et al., 2018). Furthermore, research on verbal and non-verbal rhythm processing and the timing system has made big advances in finding out about the neurological underpinnings of those abilites. It will become very obvious why at the moment the hypothesis of an internal timing deficit is one of the most plausible ones, since there is a great overlap in the neural structures underlying timing functions and the ones relevant for speech perception and production (Etchell, Ryan, Martin, Johnson, & Sowman, 2016).

Perception and production of audible rhythms engage a network consisting of sub-cortical and cortical areas, as chapter 2.6.2 has shown: The basal ganglia (BG), the SMA (Kotz & Schwartze, 2011) and other premotor cortices as well as the auditory cortex and the cerebellum (CB) are involved in perceiving, processing and producing rhythm (J. L. Chen, Penhune, & Zatorre, 2008; J. L. Chen, Zatorre, & Penhune, 2006; P. A. Lewis, Wing, Pope, P., & Miall, 2004; Schwartze & Kotz, 2016; Zatorre, Chen, & Penhune, 2007). One region in which many studies found differences in adult PWS is the BG (Alm, 2004; Chang & Zhu, 2013; Civier et al., 2013; Giraud et al., 2008) and the CB (S. Brown et al., 2005) during different verbal tasks. These areas are relevant for the mediation of temporal information and the coordination of the suitable motor responses to it but also for the sensorimotor integration (Kotz & Schwartze, 2010; Wing, 2002; Zatorre et al., 2007). Within the CB, reduced connectivity of the three cerebellar peduncules was found in PWS compared to PWNS and a hyperactivation of the cerebellar vermis was detected (S. Brown et al., 2005; Connally et al., 2014). These results go in line with many further studies that showed differences in the basal ganglia-thalamocortical circuit for adult PWS (Fox et al., 2000; Ingham et al., 2004; Lu et al., 2010; K. E. Watkins et al., 2008). But also in the connection of the BG and the premotor area an aberrant connectivity in PWS was found during tasks that involved speech planning, whereas in tasks involving speech production the cerebellar-premotor circuit was affected (Lu et al., 2010). Additionally, a positive correlation between stuttering severity and the activation patterns of the caudate nucleus was found as well as a negative correlation with activation in the substantia nigra of the left hemisphere by Giraud and colleagues (2008). But not only on the subcortical level, also in cortical areas differences between PWS and PWNS have been found, overlapping again with the motor timing circuit and areas relevant for sensorimotor integration. This applies for the SMA (Kotz & Schwartze, 2011) in which a lower amplitude of low-frequency fluctuations was found in a MRI study conducted with PWS and PWNS (Xuan et al., 2012), whereas during speech production in PWS exactly the same area showed a hyperactivation (S. Brown et al., 2005). In the left ventral PMC, an area of great interest, since it is associated with the planning of articulatory movements and the integration of motor actions and following sensory consequences, white fiber tracts displayed a reduced integrity in PWS (Civier et al., 2013; Kohler et al., 2002; K. E. Watkins et al., 2008; Wise, Greene, Büchel, & Scott, 1999). Although those results give us a better insight into the aberrant ways of processing and producing speech-relevant information in PWS, we are still left in great uncertainty about the actual causes of stuttering. Since nearly all studies focusing on structural and functional brain anomalies in PWS were conducted with adult stutterers and only very few with children, it is hard to differentiate between anomalies in structure or function that are of causal and those which are of compensatory nature.

One study that has been cited before in chapter 2.5.2.2 brings very relevant information to this issue: Sowman and colleagues (2014) compared patterns of brain activity measured by magnetoencephalography of CWS and CWNS during a picture naming test. Results show no group differences between CWS and CWNS in matter of brain activity and its lateralization, which could support the hypothesis that the aberrant lateralization of brain function in adult PWS may rather be a result of neuroplasticity due to compensatory processes as stuttering becomes chronic, than an actual cause of stuttering itself (Sowman et al., 2014). In contrast, Chang and Zhu (2013) compared functional connectivity in CWS and CWNS aged three to nine years and showed that already at this young age there is a reduced connectivity between the putamen and the SMA, the STG and the CB as well as between the SMA and the putamen, STG and CB. Therefore, the authors concluded that CWS in comparison to CWNS show reduced and altered activity in those areas generating self-paced movement (Chang & Zhu, 2013). Corresponding results were shown in a Voxel-Based-Morphometry study which was also conducted with children who do and do not stutter (Beal et al., 2013). In this study they compared the volume of the caudate of boys who stutter and a fluently speaking control group. Since they found out that boys who stuttered had significantly less volume in the right caudate compared to the controls they came to the conclusion that, even at such young age, CWS show aberrant structures and connectivity in the internal timing network (Foundas et al., 2013). A white matter tractography focusing on the neural network for speech-motor control in CWS found higher fractional anisotropy and axial diffusivity in the right frontal aslant tract, a structure relevant for the connection of motor and pre-SMAs within the superior frontal gyrus. For the first time, authors found a higher density of the frontal aslant tract in CWS compared to CWNS in the right hemisphere, hinting towards a high myelination and integrity of those tracts in young stuttering participants. Those results could indicate the emerging compensation of the right hemisphere in young CWS as the left hemisphere shows subtle aberrations in the motor-control network. Overall, those studies conducted with children support the contention that stuttering is a condition caused by neuroanatomical and neurofunctional aberrations, especially in the BG, although those aberrations may not be quite as prominent as they are in adult PWS (Etchell et al., 2014).

Besides those neurological underpinnings, articulatory theories of stuttering seem relevant for this chapter especially since stuttering is thought of as a problem of sensorimotor integration. Timing variability, as studies have demonstrated, is a signature of stuttering when it comes to tasks of rhythmic synchronization in motor behavior such as finger-tapping or clapping. Articulatory parameters however, also seem affected by this timing deficit, as various studies implicate: One very interesting study by Max and Gracco (2005) compared the coordination of laryngeal and oral movements of PWS and PWNS during perceptually fluent speech production. Results of combined kinematic and electroglottographic analyses showed that PWS differed from PWNS even in their perceptually fluent speech in terms of duration of voiceless bilabial stops. Overall, this study's results suggest that articulatory kinematics reveal more temporal variability. Falk and colleagues (Falk et al., 2016) investigated the VOT in voiceless stops, such as /p/, /t/, or /k/ during a speaking and singing task. Results show a lower VOT variability in singing compared to speaking and more variability in utterance duration in stuttering versus non-stuttering participants, notwithstanding the above forms of vocalization. A further study that was conducted by Sares and colleagues (2018) aimed to investigate the impact of simultaneous auditory feedback in order to get closer to the differences in sensorimotor integration in stuttering. Sares and colleagues found a higher variability in the timing of their response for PWS compared to PWNS, indicating a less robust internal control system and a reduced coupling of auditory and speech motor systems. To investigate the inter-articulator coordination, Smith and colleagues (2010) conducted a study with adult PWS: They assessed participants performance on a non-word repetition task, varying the test-items in length and phonological complexity. Results of kinematic data show big differences between groups concerning the consistency of coordinative patterns, hinting towards differences in the speech motor dynamics underlying perceptually fluent speech in PWS. Adding linguistic or phonological complexity, those factors are likely to contribute to a breakdown of the speech motor system leading to stuttering symptoms. In sum, results of kinematic studies have demonstrated that PWS, even when producing perceptually fluent speech, show aberrations in articulatory movements (McClean et al., 1990; G. N. Zimmermann, 1980).

Altogether, those results show that there are temporal deficits of PWS in the verbal and the non-verbal domain, since not only is the speech production temporally more variable, but also the nonspeech movements. These findings therefore support the idea that timing mechanisms within the motor system may be a potential source of deficits resulting in stuttering, and both the verbal and non-verbal motor deficits could emerge from a common deficient timing system. Those deficient timing mechanisms can negatively affect the onward flow of speech in PWS. Some models on stuttering speech production have proposed that a core issue is the malfunctioning auditory-motor integration (Max et al., 2004; Neilson & Neilson, 1987). Harrington (1988) also addressed this issue in his model and hypothesizes that PWS have problems in predicting the vowel of stressed syllables, since their auditory feedback relies on a malfunctioning perception one can describe as anticipation (see chapter 2.6.3). This way of explaining stuttering symptoms is very reasonable considering the fact that external timing or the overlayering of their own acoustic feedback is extremely helpful for PWS and can even enhance their speech fluency (Harrington, 1988; Stager, Jeffries, & Braun, 2003; Toyomura et al., 2011). Results have shown that stuttering symptoms are drastically reduced when speech production of PWS is paced by metronome tones or the speech of another person (i.e. choral singing, shadowed speech). It was hypothesized that fluency enhancing conditions help PWS to gain a better auditory-motor-system coupling (Stager et al., 2003) since they enhance the rhythmic structure of speech and facilitate temporal predictions during speech production.

Predictive timing in the verbal domain concerns the fine-grained articulatory movements that initiate a sound and form the transition to the following sound. The aims of the present study were to investigate whether predictive timing processes, i.e. the precise coordination of articulatory movements in alignment to a pacing event are altered in young and adolescent PWS, even under fluency enhancing conditions, such as paced speech. We test the hypothesis that children and adolescents who stutter show timing asynchronies in a verbal synchronization task and investigate how age influences those skills. Since SMS tasks are perfectly suited to test the abilities on predictive timing because they involve the coupling of precise motor movement with a predictable sound sequence, it was also employed in this study in form of a verbal synchronization task (Neef, Jung, et al., 2011; Repp & Su, 2013). We hypothesize that speech production to an external pacemaker - although it might lead to a more fluent speech – still shows signs of altered predictive timing in terms of lower synchronization accuracy (Falk et al., 2015) as well as higher variability. Furthermore, the differences between the experimental and the control group are expected to be most measurable when increasing the difficulty by using different synchronization tempi or using more complex stimuli. To investigate the link between disruptive predictive timing and syllabic structure or, more precisely, if an incorrect prediction of the syllabic rhythmic event is associated with atypical timing in the transition from syllable-initial consonants to its following vowels, stimulus material was constructed to control for syllabic structure and for the type of segmental material.

4.2 Material and methods

4.2.1 Participants

54 native German-speaking children and adolescents displaying developmental stuttering participated in the experiment. Due to comorbidities (dyslexia, ADHD, and cluttering) or cognitive impairment (learning disability), 14 participants who stutter were excluded from the analyses. The final sample consisted of 40 PWS (5 females, 35 males) with a mean age of *M* = 12.86 years (*SD* = 2.49) and a randomly selected control group of 40 age- and gender-matched children and adolescents (*M*= 12.71 years, *SD* = 2.47). PWS were recruited and tested on general measures and speech-rhythm-tasks (see chapter 4.2.2) prior to a therapy course held near Munich (staerker-als-stottern.de). Stuttering severity was assessed by trained speech therapists using the SSI-3 (Riley, 1994) and the German version of the OASES (Yaruss & Quesal, 2006), with the first being an objective measure to quantify symptoms and the latter being an assessment of the subjective psychosocial impact of stuttering on the everyday life of a person who stutters. The SSI-3 scores ranged from very mild to very severe stuttering with a mean moderate severity of *M* = 27.08 (*SD* = 10.73), the scores for the psychosocial impact ranged from very mild to moderate-severe with a mean moderate impact of *M* = 2.37 (*SD* = 0.51) (see Table 4.1).

Severity	**SSI-3**		**OASES**	
category	**n**	**%**	**n**	**%**
1	2	5	1	2.5
2	8	20	16	40
3	10	25	19	47.5
4	11	27.5	4	10
5	9	22.5	0	0

Table 4.1 Distribution of participants per severity category for SSI-3 and OASES (*n* = 40)
SSI-3: 1 = very mild; 2 = mild; 3 = moderate, 4 = severe; 5 = very severe;
OASES: 1 = mild; 2 = mild-moderate; 3 = moderate; 4 = moderate-severe; 5 = severe

As rhythmic capacities may be influenced by musical training, working memory capacity and inhibitory control, we assessed whether groups were comparable on these measures. In the digit span, a subtest of the Wechsler Intelligence Scale for Children (Wechsler, 2010), PWS achieved a mean of 9.63 (*SD* = 3.03) and showed a performance comparable to the age-matched control (*M*= 9.40, *SD* = 1.97). Furthermore they participated in a Go-Nogo Paradigm, a subtest taken from the TAP-Battery (P. Zimmermann & Fimm, 2002), where results also showed a comparable performance regarding the error-percentiles, which are a good way of measuring inhibitory control. The group of PWS had a mean of 50.15 error percentiles (*SD* = 32.04) on this task and with a mean of *M* = 53.17 (*SD* = 31.03) the control group performed similarly (see Table 4.2 for more details on participants). None of these measures differed significantly between groups and all parameters showed a homogenous distribution of variances - besides musical training, which differed significantly between groups with PWS (*M*= 1.99, *SD* = 2.51) displaying a significantly longer musical training than PWNS (*M*= 1.02, *SD* = 1.16; *t*(55) = 2.237, *p* = .029). A large span and some extreme values within the distribution of these values, especially within PWS, leads to large values for the standard deviation (SD) as can be seen in table 4.2. This fact, however, must be kept in mind when conducting analyses and interpreting results, since musical training was demonstrated to be a strongly influencing factor in synchronization tasks (Repp, 2010).

<table>
<tr><th></th><th colspan="3">PWS</th><th colspan="3">PWNS</th></tr>
<tr><th></th><th colspan="2">mean</th><th>SD</th><th colspan="2">mean</th><th>SD</th></tr>
<tr><td>Age
(in years)</td><td colspan="2">12.86</td><td>2.49</td><td colspan="2">12.71</td><td>2.47</td></tr>
<tr><td>Musical training
(in years)</td><td colspan="2">1.99</td><td>2.51</td><td colspan="2">1.02</td><td>1.16</td></tr>
<tr><td>Inhibitory control
(Error Percentiles of Go-Nogo task, TAP Battery, P. Zimmermann & Fimm, 2002)</td><td colspan="2">50.15</td><td>32.04</td><td colspan="2">53.17</td><td>31.03</td></tr>
<tr><td>Working memory
(digit span, Wechsler, 2010)</td><td colspan="2">9.63</td><td>3.03</td><td colspan="2">9.40</td><td>1.97</td></tr>
<tr><td>Education
(school type)</td><td>School type</td><td>n</td><td>%</td><td>School type</td><td>n</td><td>%</td></tr>
<tr><td>1= Primary School</td><td>1</td><td>13</td><td>32.5</td><td>1</td><td>12</td><td>30</td></tr>
<tr><td>2= Intermediate period (HS)</td><td>2</td><td>6</td><td>15</td><td>2</td><td>17</td><td>42.5</td></tr>
<tr><td>3= Intermediate period (RS)</td><td>3</td><td>8</td><td>20</td><td>3</td><td>7</td><td>17.5</td></tr>
<tr><td>4= Grammar School</td><td>4</td><td>13</td><td>32.5</td><td>4</td><td>4</td><td>10</td></tr>
<tr><td>Sex</td><td colspan="3">female: n = 5
male: n = 35</td><td colspan="3">female: n =5
male: n = 35</td></tr>
</table>

Table 4.2 Mean values and SD of PWS (*n* = 40) and PWNS (*n* = 40) on general measures

To explore potential differences in developmental trajectory, each participant was assigned to either a younger (9-13.25 years, “young”) or an older age-group (13.75 -17 years, “old”). Since the terms of “children” and “adolescents” do not reflect the age spans correctly, those terms were not used when talking about age groups. There were 22 PWS and PWNS each in the young group (3 female, 19 male) and 18 in the old group (2 female, 16 male). Concerning the parameters mentioned above (musical training, working memory and inhibitory control) homogeneity of variances was given (apart from musical training) and no significant differences between age groups could be found, indicating no influence of age on these measures (working memory: $t(78) = -.136, p = .892$; inhibitory control: $t(76) = -.309, p = .758$). Also in musical training, a Welch t-test confirmed that there were no significant differences between age groups with $t(57.55) = -.951$, $p = .345$. This information is relevant when comparing performances on speech synchronization tasks, since all potential differences found are to be linked to synchronizing ability developing with age, but not to general differences on cognitive measures between age groups. Furthermore, stuttering severity and the impact of stuttering displayed homogeneity of variances as well and showed no significant differences between age groups (all $p > .05$). Again, this fact must be kept in mind when interpreting results on speech synchronization tasks, since any found differences in synchronization performance are merely to be ascribed to age and not differences in stuttering severity. Since no significant differences between the two age groups were found regarding any of the variables tested, the two age groups can be merged or analyzed separately, depending on the questions raised.

Participants and their parents were informed prior to the study and gave informed consent. The study was approved for conformity with ethical standards by the ethics committee of the faculty of medicine of the Ludwig-Maximilians-University Munich.

4.2.2 Stimuli and procedure

For the targeting of alignment skills in the verbal domain, participants were asked to utter four different types of stimuli: nonsense syllables, with either simple (“ba”), or complex (“bla”) onsets and monosyllabic words also with either simple (e.g. “Lauch”) or complex (e.g. “Schleim”) onsets. The word lists contained 24 monosyllabic words each (12 nouns and 12 adjectives, see Appendix 2), with one list exclusively containing words with simple onsets and the other containing words with complex onsets. Simple words started with the phonemes /t/, /k/, /S/, and /l/ with 6 words (3 nouns, 3 adjectives) for each phoneme. Complex onsets comprised the phoneme combinations /kl/, /Sl/, /St/ and /bl/ (3 nouns and 3 adjectives for each phoneme combination). Low-frequency words and abstract nouns were excluded. Frequency counts (retrieved from SUBTLEX-DE; (Brysbaert et al., 2011) for simple (mean = 2.75 (log10 frequency), SD = .83) versus complex (mean = 2.84 (log10 frequency), SD = .56) word lists did not differ ($p > .66$). For the paced reading task, the word lists were arranged in written form on a DIN-A4 sheet of paper (Arial, 14, landscape format), preceded by a practice trial of 5 words for each list. The selection of these stimuli was based on the fact that timing could also depend on syllable complexity and a simple consonant-vowel-structure might demand less articulatory planning and execution capacities than a complex CCV-structure as previous results have shown so for phonological complexity (Smith et al., 2010). Furthermore, this comparison might give a better insight into ongoing processes of articulation in stuttering speakers about their internal timing as data can reveal if aberrations occur only rather in form of a delay or if it is the transition from consonant to vowel resulting in timing asynchronies.

During the unpaced condition, participants were asked to repeat the syllables and subsequently to read the words aloud at a self-chosen and comfortable pace. In addition, participants were instructed to speak as steadily and evenly as possible (i.e. not to accelerate or slow down) and were given the opportunity to familiarize themselves with the word list prior to the recording. The order of stimulus type (simple versus complex onsets) was counterbalanced across subjects. After the unpaced trials the actual synchronization task begun: Participants were asked to synchronize their own productions of syllables and words to “beeps” of the metronome. The metronome rate was 750ms IOI for syllables, and 900ms IOI for the wordlists. The specific instruction was to read each syllable/word to one “beep” of the metronome, starting whenever they felt ready and familiar with the rhythm. The first 5 syllables/words of a trial were counted as a training phase. If participants did not succeed in synchronizing their productions after the “training phase” (i.e. severe misreading or interruptions caused by stuttering), the trial was restarted. The metronome was presented in the free field via Neusonic NE03 studio monitors installed in front of the participant at a comfortable sound pressure level. Metronome beeps were recorded with a microphone installed in front of the studio monitors. Participants’ speech was recorded with a Beyerdynamic condenser microphone (Type TG H54c) contained in a comfortable head-set, which was connected to the ZOOM H4n-Handy Recorder. Metronome and speech were hence recorded simultaneously with separate channels of the ZOOM Recorder. One session lasted approximately 45 minutes. The experimenter was present during the whole session.

4.2.3 Data preparation and analyses

Prior to all main statistical analysis, data of the verbal synchronization task had to be edited. Throughout all conditions - paced and unpaced - speech data were segmented and syllable and vowel onsets of the fluently spoken words were marked and edited by inspecting the oscillogram and spectogram of the audio signal in Praat (Boersma & Weenink, 2009): words and syllables were segmented in an orthographical and phonetic (only boundaries of words and the beginning and end of the target vowel) segmentation, with clear rules for boundaries. Pauses were segmented as <*p:*>, or *Atmen* <*p:*> if a respiration was audible. If participants misread or misspoke a word, it was excluded from the analyzes by marking it with *, syllables containing stuttering symptoms were also excluded and marked with #. The decision about the presence or absence of a stuttering symptom was done by a trained speech therapist specialized in stuttering. All remaining words and syllables were segmented with the following guidelines: bursts of plosives were marked by inspecting the oscillogram, vowel onsets were segmented at the second zero-crossing of the first clearly visible period. Diphthongs were treated as one segment. The end of a vowel was defined as a missing of clear formants, frications were not classified as part of a vowel. For the segmentation of the "/l/" in words or the syllable "bla" the first positive zero-crossing of the period was marked as the beginning. In the case of a plosive occurring in the middle of a word, the time between burst and voicing was added to the consonant. The segmentation of the individual metronome pulse recorded at the second channel, was done in the second positive zero-crossing. In analogy to p-centers, the vowel (it's onset, duration and offset) was considered the most relevant part of the syllable/word; therefore, there was no differentiation of different onsets of the words, besides the already explained one: simple and complex. All phones occurring prior to the vowel were therefore segmented as one unit. Duration of those segments could consequently vary a lot, depending on the phonemes contained within this segment.

After the segmentation of those words and syllables in *Praat*, files were extracted as textgrids for further analyses. An example of a Praat- file can be seen in figure 4.1. Using Matlab version R2017b (MATLAB, 2017), all text-files were scanned for word- and vowel onsets, durations of vowels and words as well as offsets of vowels and words.

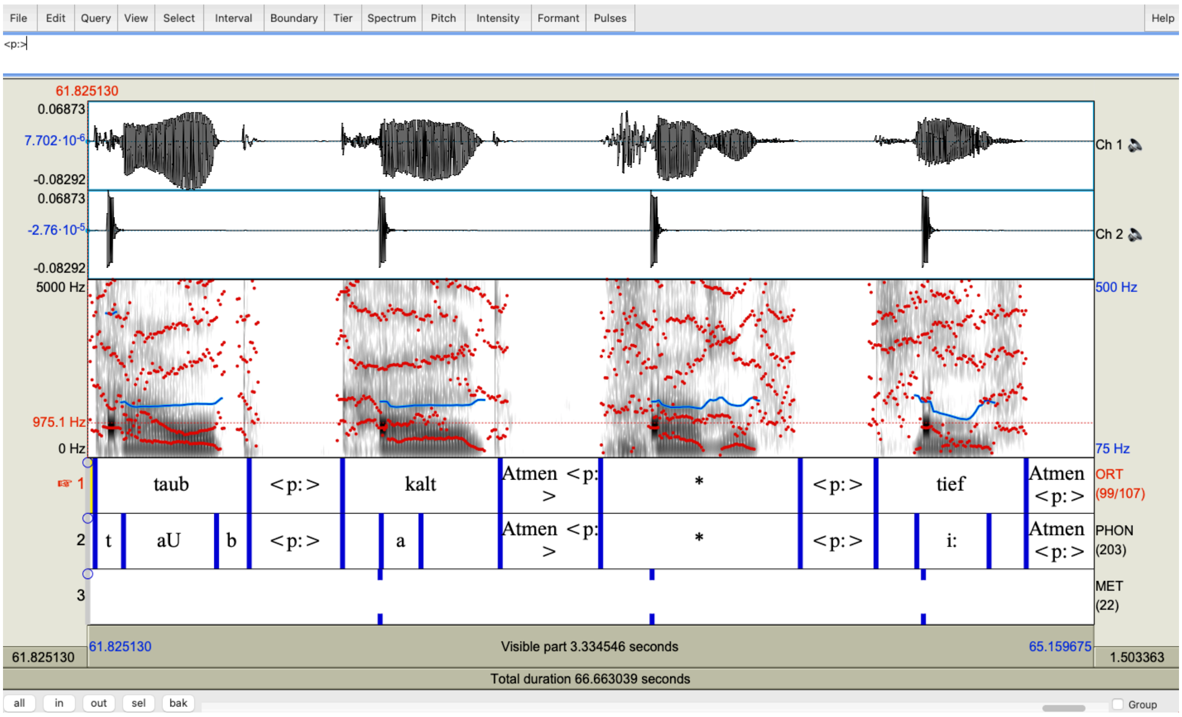


Figure 4.1 Example of a Praat-file displaying the relevant markings for the calculation of synchronization parameters

For unpaced speech, the mean inter-vowel-interval (IVI) of syllables/words was analyzed to get information on spontaneously chosen mean intervals of synchronization. Furthermore, the Coefficient of Variation (CV; i.e. variability of the inter-vowel-intervals) was analyzed by calculating the ratio of the standard deviation to the mean onset time (vowel or consonant, depending on research question). For the two paced conditions (i.e. syllables and words), synchronization consistency and accuracy were measured by evaluating the time of the vocalic and the consonantal onset of the syllable/word in relation to the metronome tone. Consistency refers to variability in the performance, i.e. how evenly onsets/vowels were produced, whereas accuracy stands for the precision with which the synchronization performances was obtained. High consistency is reached if a participant always utters the syllable/word at the same time relative to the metronome and corresponds to the length of the resultant vector *R*. This vector can reach values ranging between 0 and 1, whereby values close to 1 indicate a high consistency and therefore a very steady performance and values close to 0 indicate a rather random performance. A good accuracy is obtained when the onsets are always very close to the pacing event. It is expressed by the angle (in degree) of the vector *R* and can therefore either be negative, if a participant uttered the onset before the pacing event, or positive, if a participant uttered the onset after the pacing event. A more detailed explanation about the exact way of calculating the resultant vector R is summarized in the following section.

To this end, we used circular statistics to acoustically analyze the segmental timing in relation to the pacing event (Berens, 2009), after the raw synchronization times (concerning vowel and consonant) had been preprocessed: onsets of the first 3 syllables/words were removed as well as onsets with an IOI lying outside the 3*inter-quartile range from the median IOI. Sequences of syllable/word production were then further processed using CircStat-Software for Matlab, which has been used previously for analyzing and processing synchronization data (Berens, 2009; Falk et al., 2015; Sowinski & Dalla Bella, 2013). The advantage of circular compared to linear statistics is that the former does not require a one-to-one correspondence between the pacing event and, in this case, the verbal response (i.e. syllable or word spoken). In addition, metrics derived by it have proven to be more sensitive when uncovering individual differences in tasks of SMS (Falk et al., 2015).

Productions of vowel or consonant onsets are represented on a full circle of 360° (see figure 4.2), which indicates the IOI (750ms or 900ms) between the pacing events recurring on a regular base. Each onset is represented by an angle in relation to the pacing event (0°). The angles corresponding to the onsets where then transformed into unit vectors, of which the mean resultant vector R is computed (Berens, 2009; Fisher, 1993; Mardia & Jupp, 2000).

This vector serves to calculate the measures of synchronization performance, specifically consistency and accuracy (Sowinski & Dalla Bella, 2013). Values for accuracy were only calculated if a participant's performance was above chance, i.e. if data points were not randomly distributed around the circle; therefore, values exceeding +/-90° were removed. This was assessed with the Rayleigh test for circular uniformity (Falk et al., 2015; Fisher, 1993). Moreover, prior to the statistical analyses, values for vector length were logit-transformed to diminish data skewness. All following statistical analyses were conducted in SPSS (Version 27).

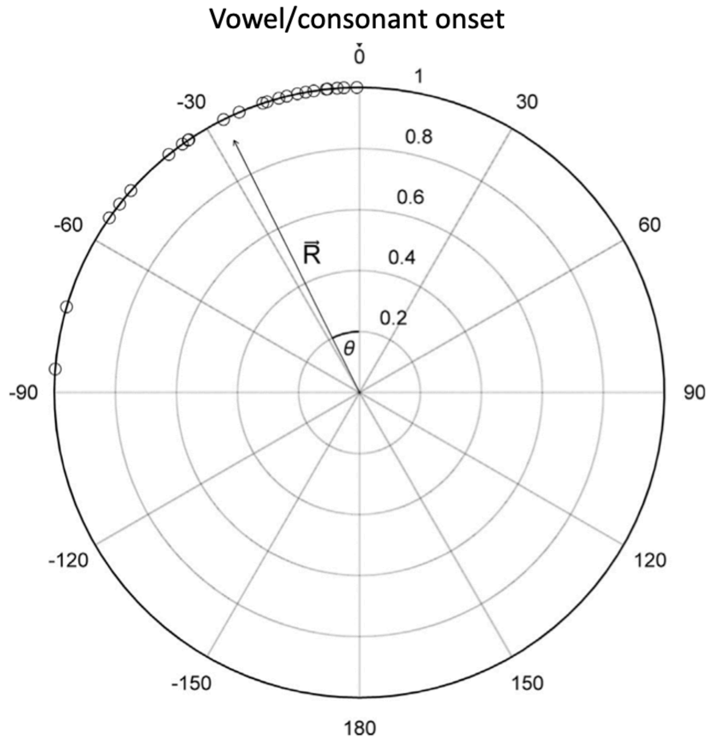


Figure 4.2 Visualization of Circular Statistics and the resultant vector *R* (adapted figure from Sowinski & Dalla Bella, 2013, p.1954):
Exemplary distribution of vowel or consonant onsets in a verbal synchronization task. The figure also shows the direction (angle theta θ) of the resultant vector *R* indicating if the synchronization took place prior to or after the pacing event. For each trial the same procedure was computed: directions were transformed into unit vectors, afterwards they were averaged in order to obtain *R*. Length of the vector therefore stands for the consistency in synchronization (the closer to 1 the value of *R* is, the more consistent the performance). For this illustration the angles are arranged to proceed clockwise from 12 o'clock.

4.3 Results

For a better understanding, results can be subdivided - in analogy to the testing procedure - into the unpaced (no external rhythm) and the paced (synchronization to a given rhythm, i.e. the metronome) condition. Furthermore, a division into inter-vowel versus inter-consonant-intervals will be made in the paced condition.

4.3.1 Unpaced condition

Prior to investigating mean IVIs and CV of the IVIs of the unpaced condition, duration times in syllables and words were compared between groups to see if there might be an influence of the mere duration of the production. To do so, t-tests were calculated on mean duration times of syllables (both simple and complex) and the vowel within a syllable as well as mean duration times of words (both simple and complex) and for the vowel within the words. Prior to those t-tests, data were checked for normal distribution and homogeneity of variances: Six Participants, namely P10, P28, P131, P138, P146 and P14 were classified as outliers due to extreme values in duration times and excluded from calculations on duration times. Duration times were approximately normally distributed for PWS and PWNS as assessed by the Shapiro-Wilk-Test, $p > .05$ for all variables. However, homogeneity of variances was only given in case of mean word and vowel duration for complex words ($p > .05$), for simple and complex syllables as well as for simple words the mean word and vowel durations lacked homogeneity of variances, as assessed by the Levene's-Test (all $p <. 05$). In case of missing homogeneity of variances, a Welch-t-test was conducted: Results of all t-tests, however, displayed significant differences between PWS and PWNS (all $p <. 05$), with PWS displaying significantly longer duration times in syllables and words compared to PWNS (mean duration PWNS: $M = .39$ sec., $SD = .06$; mean duration PWS $M = .44$ sec., $SD = .08$ with $t(70.027).= 2.892$, $p = .005$). To ensure that potential group effects in the unpaced condition were not a result of mere duration times in syllable/word production, all analyses were additionally run with mean word production times (averaged over all conditions) as a covariate.

In the unpaced condition, IVIs of the syllables spoken and words read were measured as well as their variability as expressed in the coefficient of variation (CV IVI). Those data were entered in a 2 x 2 x 2 x 2 mixed factorial Analysis of Variance (ANCOVA) with the between subject factors group (PWS versus PWNS) and age group (young versus old) and the within subject factors stimulus material (syllables versus words) and syllabic complexity (simple versus complex onsets). Due to the significant differences found between groups on mean duration times an ANCOVA was chosen to account for the significant differences in terms of mean duration times between groups. Musical training, which also differed significantly between groups was not added as a further covariate, though, since correlations did not display any significant influence of it on unpaced tasks (all $p > .05$). Three participants (P51, P70, P78) were excluded from further analysis on unpaced performance due to extreme values caused by severe stuttering symptoms.

Prior to detailed analyses on IVIs a visual overview shall be given to allow a better understanding of the different types of stimuli used and effects to be expected: as figure 4.3 illustrates, groups perform comparably on all types of stimuli. As expected, older participants tend to display slightly shorter IVIs, meaning that they prefer a faster tempo in the unpaced task which is true for all conditions besides the simple syllables, where old PWS even show the highest variability concerning the mean IVI. A further finding is the fact that for words the mean IVI is slightly smaller as opposed to syllables.

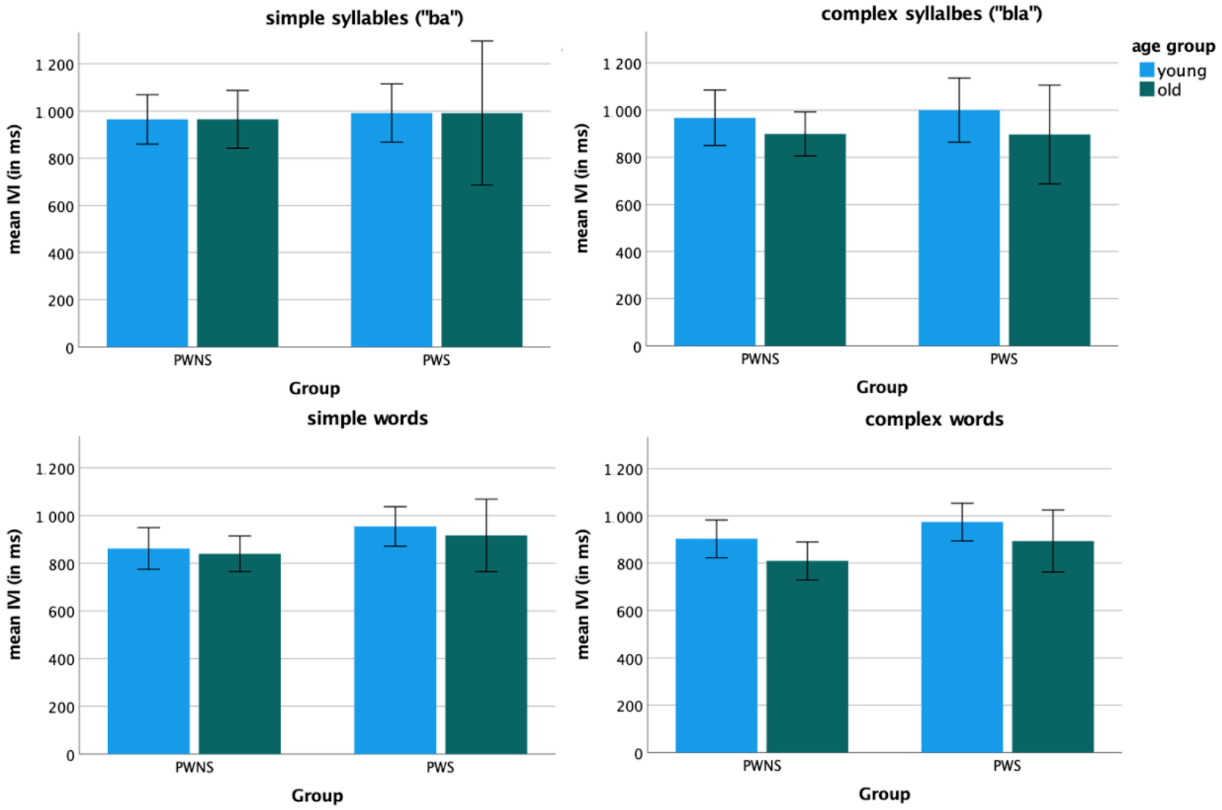


Figure 4.3 Mean IVI for each condition.
Error bars represent 95% confidence interval.

Data on **IVIs** in both groups (PWS and PWNS) and age groups (young and old) in the unpaced condition showed normal distribution as assessed with the Shapiro-Wilk-Test (all p > .05); only within the older group and the task of simple words did the data lack normal distribution (p < .05). Sphericity was controlled for with the Mauchly-Test. There was homogeneity of variances, as assessed by Leven's test (*p* > .05) for all four chosen variables. Results of the ANCOVA displayed no significant main effect for group or age group as well as no significant effect of stimulus material (syllable versus word) or any interactions.

When visualizing those results (see figure 4.4), tendencies become visible, especially when looking at the more difficult stimuli (i.e. words). Here it becomes evident that PWS display larger IVIs than PWNS and that young participants - compared to older ones - also display larger IVIs. These results hint towards a slower articulatory process in groups confronted with higher demands (i.e. stuttering participants and younger participants). However, results failed to reach significance and can therefore only be seen as mere tendencies.

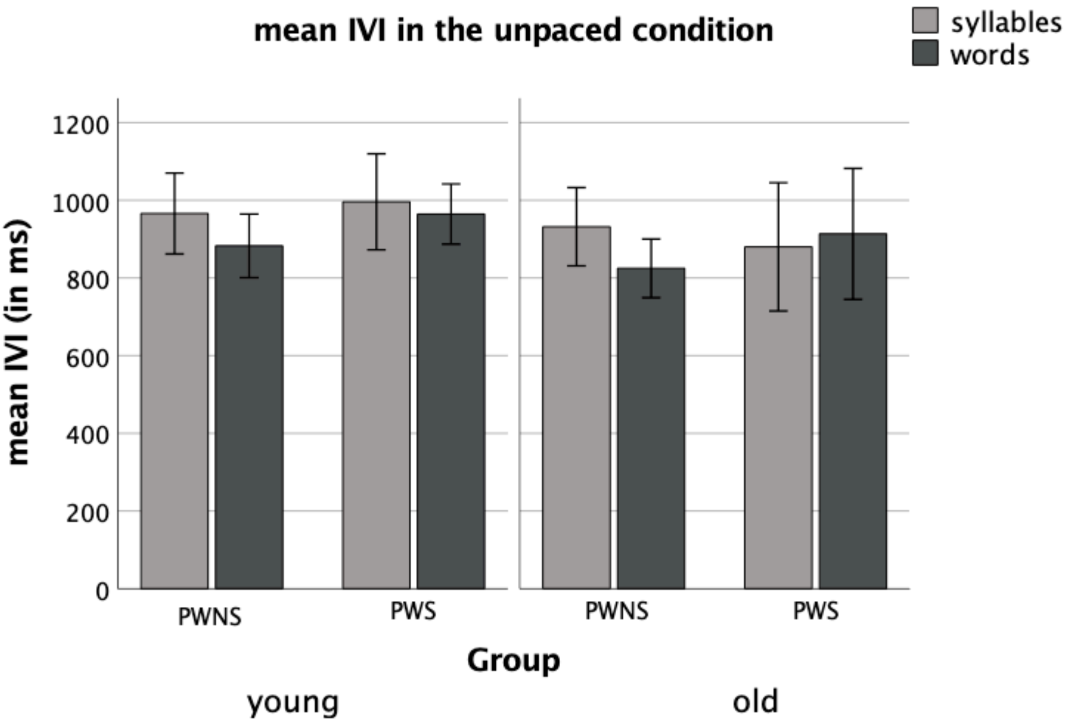


Figure 4.4 Mean IVIs of syllables and words (simple and complex merged) in the unpaced condition.
Error bars represent 95% confidence interval.

Furthermore, **articulatory variability** (CV of IVI) was analyzed: Prior to those analyses, 1 participant from the group of PWS and 3 participants from the group of PWNS had to be excluded due to extreme values, namely P80 and P147, P148 and P162. Normal distribution was checked using the Shapiro-Wilk-Test: All variables lacked normal distribution even after removing participants with extreme values. Sphericity was controlled for with the Mauchly-Test. Due to a lacking homogeneity of variances for the CV of the IOI of the complex wordlist, non-parametric comparisons were added to the results of the ANCOVA.

Prior to any further calculations, a visualization of the CV of the IVI for all four conditions shall be given for a better understanding and a first impression of results. As figure 4.5 displays groups show quite similar patterns for complex syllables and complex words, for simple syllables, however, a huge variability becomes evident for old PWS – despite the removal of outliers. This effect might be caused by the more severe symptoms within this subgroup of PWS. Still, it raises the question why this effect is so prominent in simple syllables but not in other stimuli. However, those graphs might explain the lacking normal distribution. Furthermore, for simple words PWS in both age groups show a higher variability as opposed to PWNS with no differences evident between young and old PWS. The performance on complex syllables is nearly identical for both groups and age groups, for complex words the trend is also comparable, with only young PWS displaying a rather big variability.

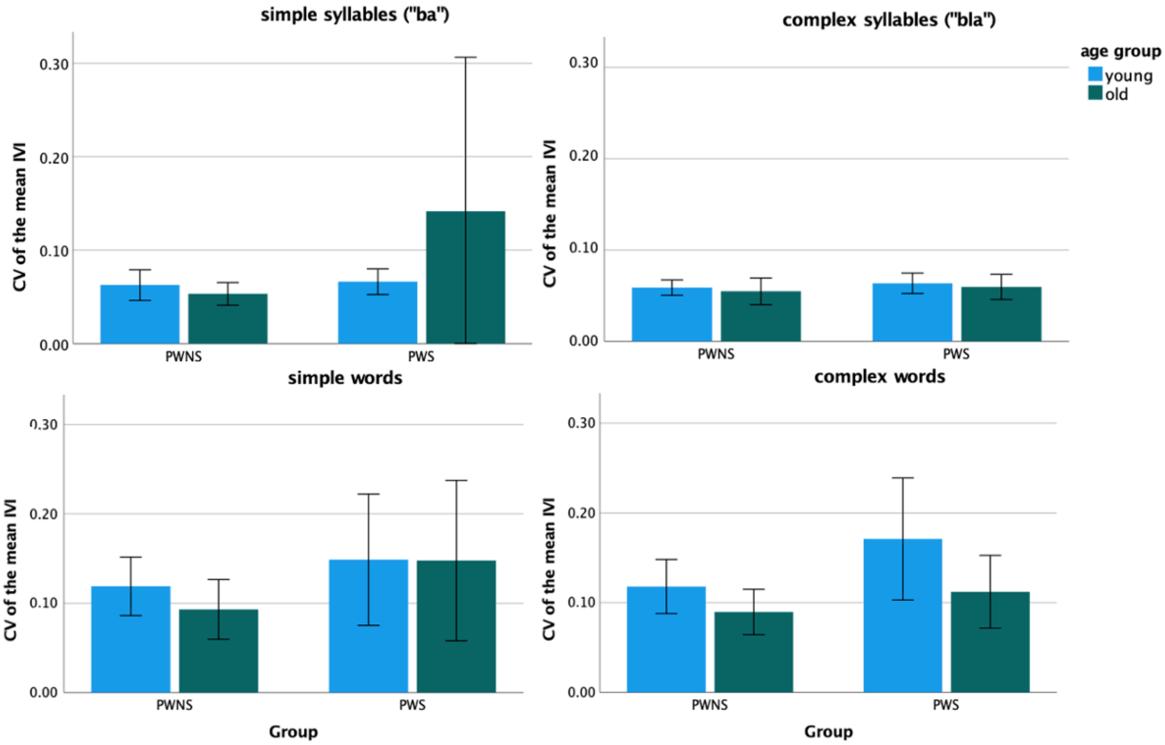


Figure 4.5 CV of the mean IVI for each condition.
Error bars represent 95% confidence interval.

After the visual inspection of all conditions for the CV of the IVI, results of the analyses are presented: neither for age-group, nor for group, can a significant difference for the articulatory variability be reported. As figure 4.6 illustrates, a significant effect of stimulus material (syllables versus words) was found with words displaying a significantly higher articulatory variability as shown with the ANCOVA ($F(1, 69) = 6.084$, $p = .016$, $\eta^2 = .081$) as well as the Wilcoxon-test ($z = -6.815$, $p < .001$). A significant interaction of stimulus material (syllable versus word) and age group was found with ($F(1, 69) = 7.026$, $p = .010$, $\eta^2 = .092$). Resolving this interaction with the help of t-tests, significant results were lost (all $p > .05$), the explorative description of mean values, though, hinted towards the fact that for syllables, the difference between young and old participants was only marginal (mean $CV_{young} = .062$, mean $CV_{old} = .056$), when looking at CVs for words, however, the difference between age groups is rather big (mean $CV_{young} = .140$, mean $CV_{old} = .108$). This effect is clearly visible in figure 4.5 with syllables displaying comparable CVs for group and for age group, whereas words display a higher variability, especially in young participants. No further effects or interactions are to be reported.

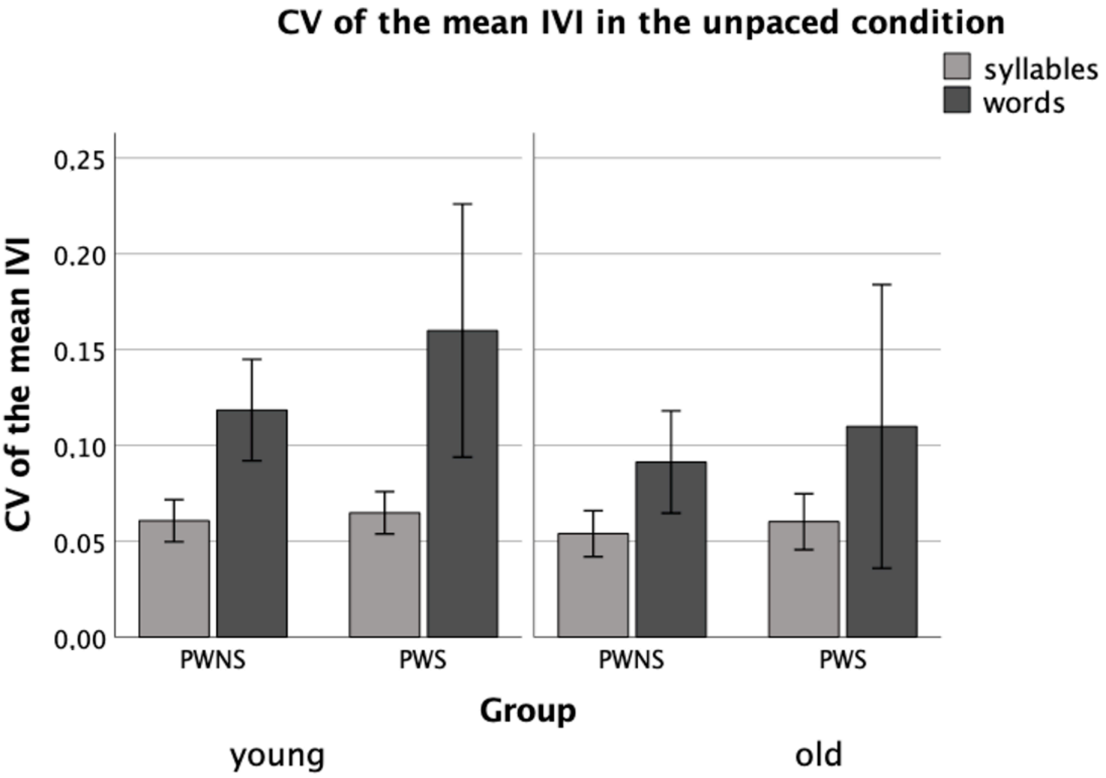


Figure 4.6 CV of the mean IVI of syllables and words (simple and complex merged) in the unpaced condition.
Error bars represent 95% confidence interval.

Those results imply that within the unpaced condition, no major differences in speaking syllables/reading words can be found between PWS and PWNS or young and old participants. Tendencies as visualized by bar plots on mean IVI and CV of IVI can hint towards trends. However, for an unpaced condition as such, mechanisms of speech production seem to work comparably. The higher variation regarding the IVI of words as opposed to syllables might be explained with the higher complexity, as onsets differ whereas syllables always remained the same. The articulation of alternating words, no matter if their onset was simple or complex, seems to consume more time during the process of articulation, as it surely is more difficult to alternate between constantly changing articulator patterns. Consequently, it seems quite logic that the IVI of syllables (that are reoccurring monotonously) is less variable compared to the IVI of alternating words. To investigate possible changes and differences in a paced speech paradigm, results of the paced conditions will be presented in the following section.

4.3.2 Paced condition

Before analyzing verbal synchronization abilities in terms of accuracy and consistency the effect of reduced motor variability due to external pacing will be investigated. As pacing is known for the stabilizing effect on motor variability, differences between CV of IVIs for the unpaced and paced condition will be compared. Although analyses on unpaced CVs did not differ between groups, it still might be possible that PWS or PWNS will benefit more from external pacing regarding their variability.

To calculate difference scores, paced CV of the IVIs were subtracted from those of the unpaced condition for each condition (syllables and words, each simple and complex). Across all four conditions, 75 % of all PWNS and 62.9 % of PWS showed a reduction of variability, 15 % of PWNS and 17.1 % of PWS showed no change in variability (i.e. a value of 0 as difference score) and 10 % of PWNS as well as 20 % of PWS showed even a more variable performance in the paced condition. In total number it is 12 Participants (6 PWNS; 6 PWS) who showed no difference in variability, 52 displayed less variability in the paced condition (30 PWNS, 22 PWS) and 11 Participants displayed a higher variability in the paced condition (4 PWNS, 7 PWS) 5 Participants (6.3 %) could not be included within these analyses due to missing values. Figure 4.7 illustrates those changes within CV of the IVI from unpaced to paced condition.

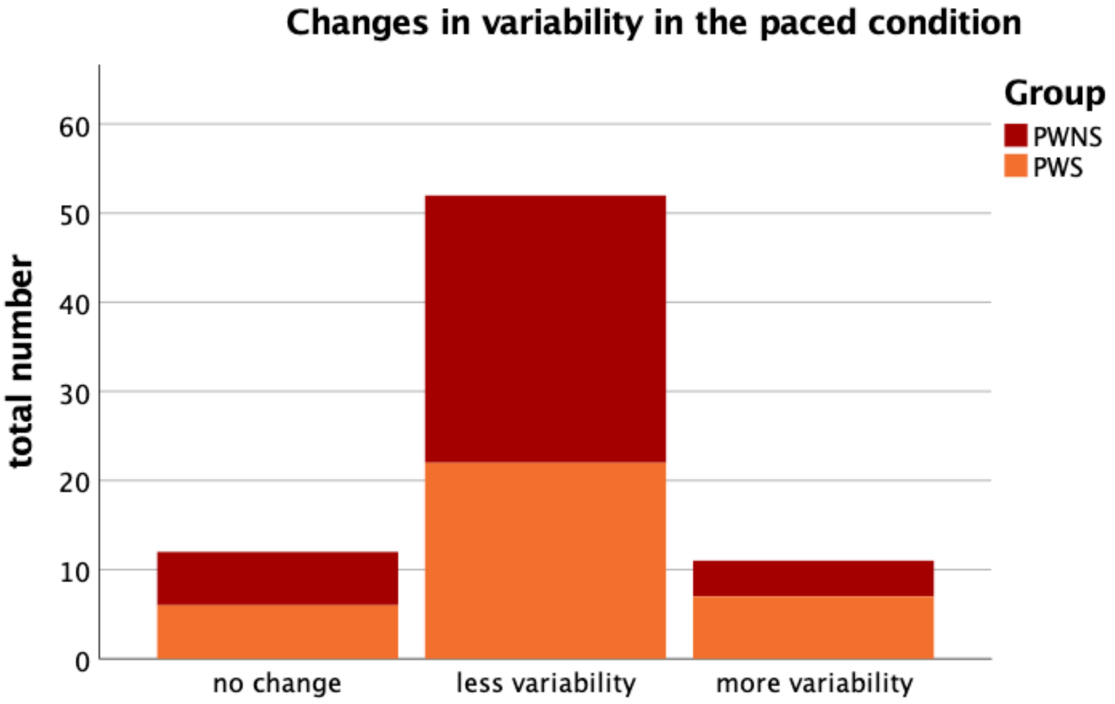


Figure 4.7 Histogram displaying changes in variability comparing paced and unpaced condition

A chi-square-test for association was conducted between groups and variability outcome. All expected cell frequencies were greater than 5. However, there was no significant difference between groups as to participants who would show a reduction of variability, a rise in variability or no effect of the condition as assessed with χ^2 (1, 75) = 1.723, p = .422, φ = .422.

Additionally, prior to investigating accuracy and consistency of the verbal synchronization in the metronome condition, duration times in syllables and words were compared to see if there might be an influence of the mere duration of the production. To do so, t-tests were calculated on mean duration times of syllables (both simple and complex) and the vowel within a syllable as well as mean duration times of words (both simple and complex) and for the vowel within the words. Prior to those t-tests, data were checked for normal distribution and homogeneity of variances: Three participants, namely P131, P138 and P28 were classified as outliers due to extreme values and excluded from calculations on duration times.

Data on duration times were approximately normally distributed for PWS and PWNS and age groups as well, as assessed by the Shapiro-Wilk-Test, $p > .05$ for all variables. However, homogeneity of variances was only given in case of word and vowel durations for simple and complex words but not for mean word and vowel duration of simple and complex syllables, as assessed with the Levene's test. In case of missing homogeneity of variances, a Welch's t-test was used instead. However, all t-tests displayed a significant difference between PWS and PWNS (all $p < .05$) with PWS displaying significantly longer duration times in the paced condition (mean duration PWS: M = .41 sec., SD = .06; mean duration PWNS: M = .35 sec., SD = .03 with t(55.983) = . 5.308, p < .001). If speech production was generally slower in PWS than PWNS, this could impact synchronization measures (e.g., lower consistency towards a reference point induced by longer durations). To ensure that potential group effects in synchronization were not a result of tempo differences in speech production, all analyses were run with mean word production times (averaged over all conditions) as a covariate. Musical training, however, which also displayed significant differences between groups was not added as a further covariate, as correlations with measures of accuracy and consistency did not display any significant correlations (all $p > .05$) and therefore no significant influence of musical training on the paced performance is to be expected.

When analyzing synchronization abilities, it is also a matter of interest if participants would rather synchronize the vowel or syllable/word onset to the metronome. As previous research in this field has demonstrated already, participants choose the vowel onset rather than the syllable or word onset (Rathcke, Lin, Falk, & Dalla Bella, 2021). This observation in productive speech can be seen as an analogy to the perceptive concept of P-centers (Cummins & Port, 1998). To account for this issue and to get more insights into processes of speech production under fluency evoking conditions, both onsets – the vowel and the syllable/word onset was used as references in calculations and compared afterwards. When discussing accuracy in terms of verbal synchronization abilities in case of this study, accuracy refers to the precision of timing the vowel or the onset of the syllable/word to the pacing event. However, accuracy must not be mistaken for the synchronization at the vowel onset. The exact point of synchronization lies close to the vowel onset; still, small deviations may occur.

Prior to detailed analyses on accuracy data for the vowel, all values exceeding 90° were removed as they were classified as outliers indicating a rather random performance instead of a real synchronization. An inspection of graphs for each condition shall help understanding this complex data better and give an overview of what to expect during the following analyses. As the inspection of figure 4.8 demonstrates, groups seem to perform comparably regarding syllables (simple and complex); when looking at words, however, groups seem to differ – at least when looking at old participants. A further fact that immediately becomes obvious is big number of outliers even though all outliers by definition had (see definition above) already been removed prior to further analyses. This hints towards the fact that in both groups this task also generated a lot of variability.

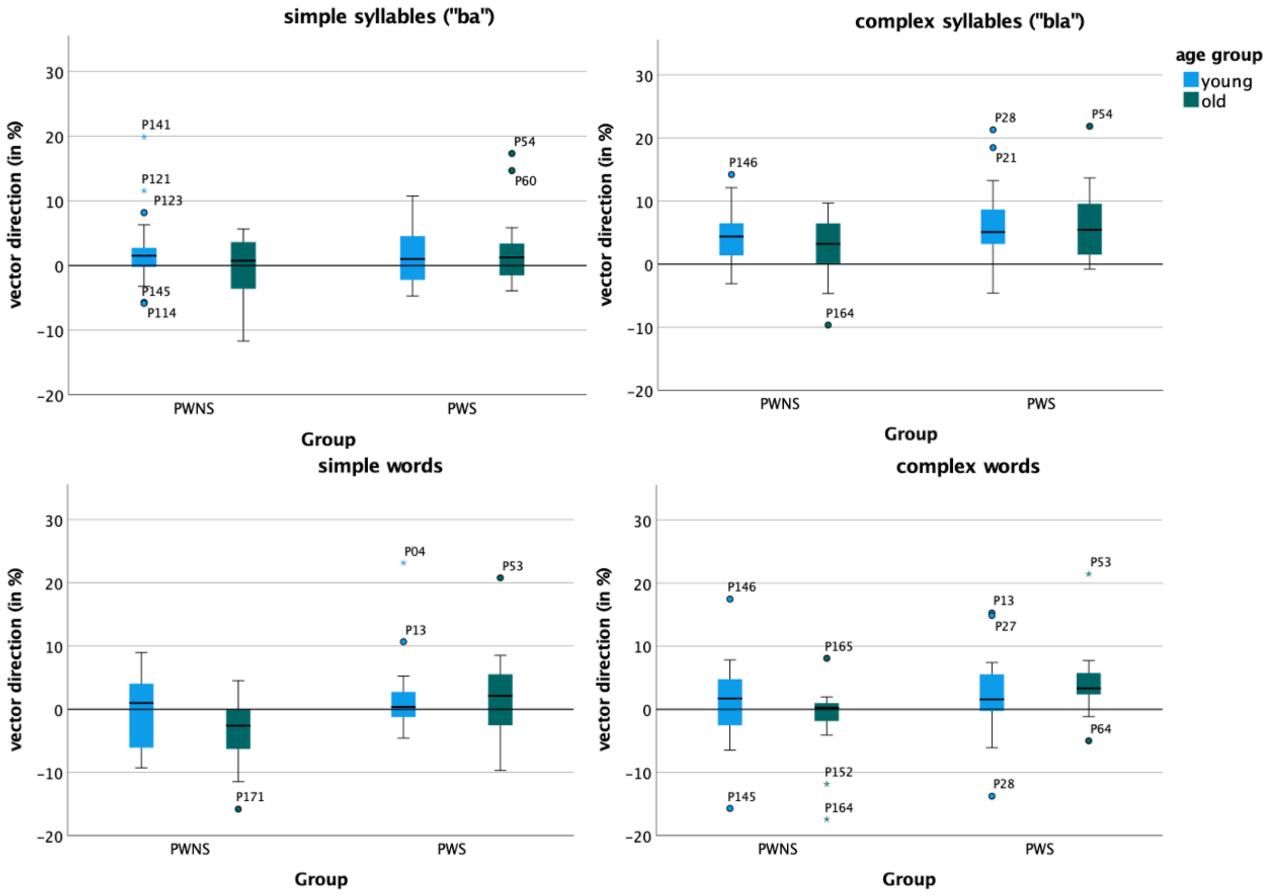


Figure 4.8 Accuracy (vector angle in % of the IVI) of synchronization for each condition (vowel as reference) in relation to the pacing event (=0).
Bars represent vector direction in both groups subdivided for age groups and display the median; details as in figure 3.5.

Accuracy and consistency data were entered in a 2 x 2 x 2 x 2 mixed factorial Analyses of Covariance (ANCOVAs) with the between subject factors Group (PWS versus PWNS) and age group (young versus old), and the within subject factors stimulus material (syllables versus words) and syllabic complexity (simple versus complex onsets). Mean duration time (paced) was added as a covariate.

Data on **Accuracy** (as measured by vector direction in % of the IVI) with the vowel as reference were controlled for normal distribution as assessed by Shapiro-Wilk, displaying normally distributed data between groups ($p < .05$) in all conditions besides simple syllables within PWS. Homogeneity of variances was given as controlled for with the Levene's test with all $p > .05$. Sphericity was controlled for with the Mauchly-Test.

The ANCOVA results show a significant main effect of group indicating a later vowel onset in PWS across conditions with $F(1,63) = 11.301$, $p = .001$, $\eta^2 = .152$ (mean vector direction$_{PWS}$ = 2.040%, SD = 0.581%; mean vector direction$_{PWDS}$ = 0.291%, SD = 0.727%, see figure 4.8). It can therefore be stated that PWS produced the vowel significantly later relative to the metronome compared to the control group. No further significant effects or interaction can be reported.

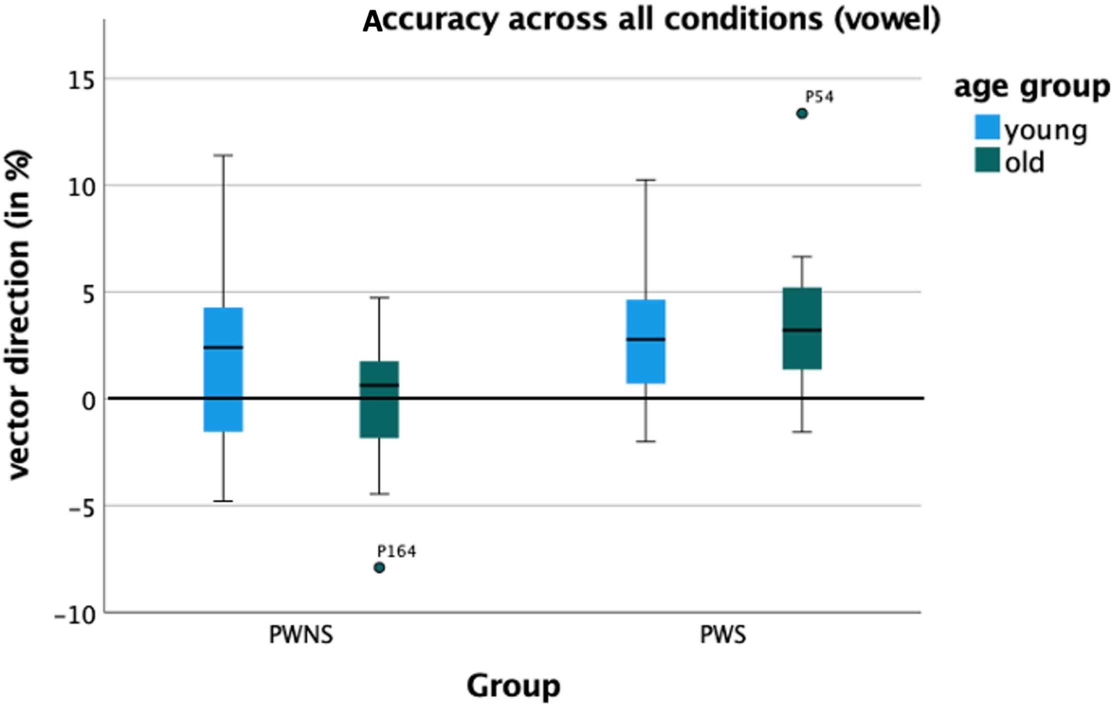


Figure 4.9 Accuracy (vector angle in % of the IVI) of synchronization across all conditions (vowel as reference) in relation to the pacing event (=0).
Bars represent vector direction in both groups subdivided for age groups and display the median; details as in figure 3.5.

The difference in vowel timing in PWS may be generated by either a vowel-specific timing delay or a general delay in word production. To check for the possibility that word production onset was generally delayed in PWS compared to PWNS, we repeated the above analysis (ANCOVA) and took as dependent variable accuracy measured with the syllable/word onset as a reference point. Prior to the ANCOVA, onset accuracy data were checked for normal distribution and homogeneity of variances: they were approximately normally distributed for PWS and PWNS as well as for young and old participants as assessed by the Shaprio-Wilk-Test, $p > .05$ for all variables; Levene's test also indicated a given homogeneity of variances for all variables. Sphericity was controlled for with the Mauchly-Test. Values exceeding the angle of 90° were excluded from the analyses, since they indicated a rather random reaction instead of a synchronization.

Prior to reporting detailed results of analyses, a visualization of each condition with the onset as chosen reference shall give an overview of these complex data and make comparisons with vowel synchronization possible. What is most evident when comparing each condition with the onset as reference (figure 4.10) as opposed to the vowel as reference (figure 4.8) is the anticipation (in relation to the metronome) that is clearly observable for the onset, but not for the vowel. Whereas for the onset a clearly negative vector direction is observed which gets more negative as stimuli get more complex, the situation for the vowel looks different: apart from one exception (old PWNS in simple words) vector direction is positive and, on top of that, a lot closer to the pacing event compared to the graphs visualizing the onset. This again, is a clear hint towards the fact that participants in both groups rather synchronized their vowel timing to the metronome, instead of the onset. Focusing on the graphs for the onset as reference, it becomes clear that groups perform comparably, as they both tend to anticipate more with complex stimuli. Young participants display more variability compared to old participants and PWS display slightly more outliers as opposed to PWNS – besides that, both groups seem to synchronize their onset with comparable patterns to the metronome with PWS showing the tendency of a slightly later onset in synchronization.

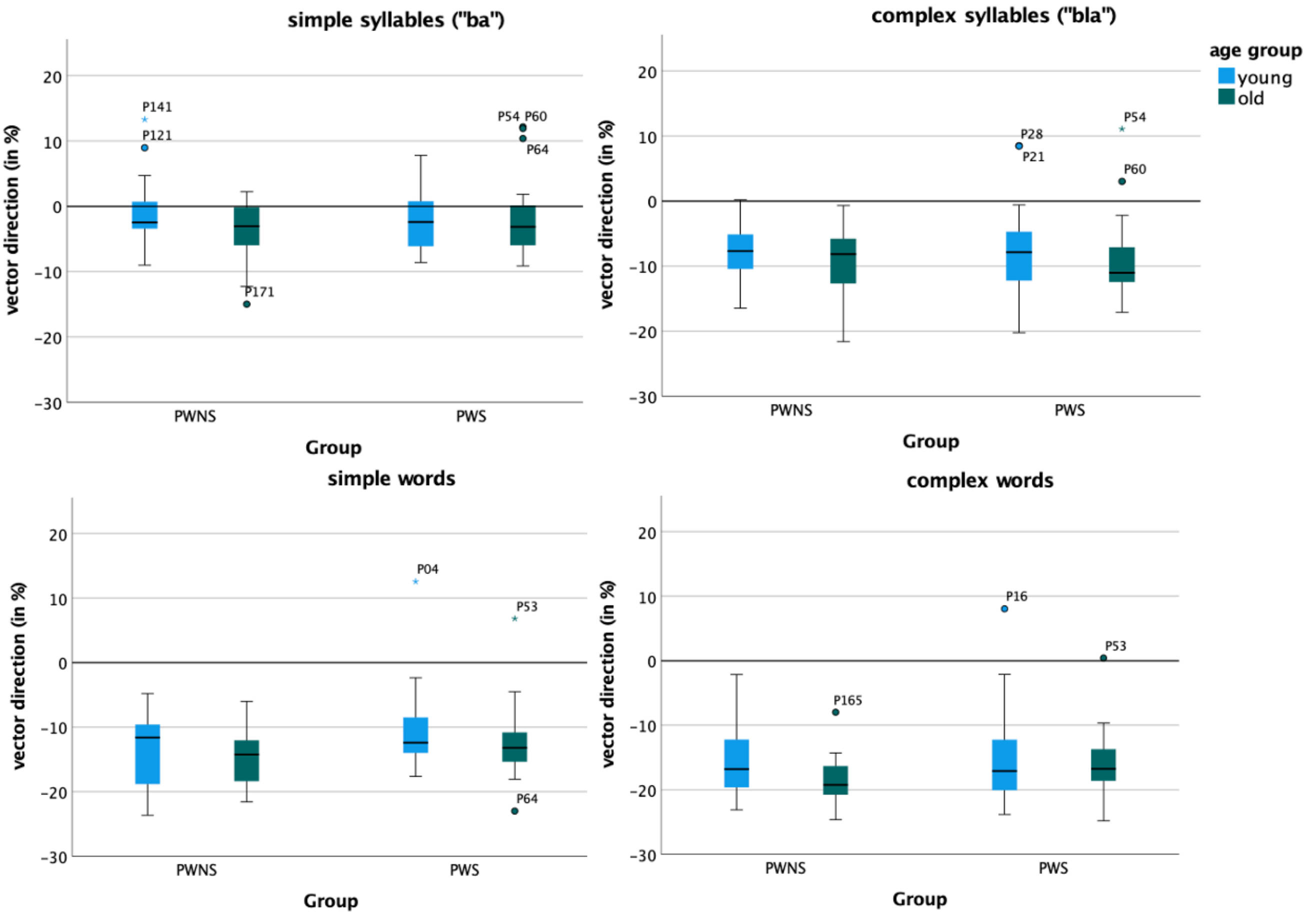


Figure 4.10 Accuracy (vector angle in % of the IOI) of synchronization for each condition (onset as reference) in relation to the pacing event (=0).
Bars represent vector direction in both groups subdivided for age groups and display the median; details as in figure 3.5.

Entering those data into an ANCOVA results show a significant main effect of group indicating a later syllable/word onset in PWS across conditions (see figure 4.11) with $F(1,60) = 5.385$, $p = .024$, $\eta^2 = .082$. It can therefore be stated that PWS do not only produce their vowel, but also their onset significantly later relative to the metronome compared to the control group (mean vector direction$_{\text{PWS}}$ = -5.404%, *SD* = 4.114%; mean vector direction$_{\text{PWDS}}$ = -10.124%, *SD* = 3.612%)

Furthermore, a significant effect of stimulus material (syllable versus word) was found ($F(1,60) = 4.423$, $p = .040$, $\eta^2 = .069$) with syllables showing a later start than words (mean vector direction$_{\text{syllables}}$ = -5.236%, *SD* = 4.934%; mean vector direction$_{\text{words}}$ = -14.424%, *SD* = 5.182%) as figure 4.9 already demonstrated. No further significant effects or interaction can be reported.

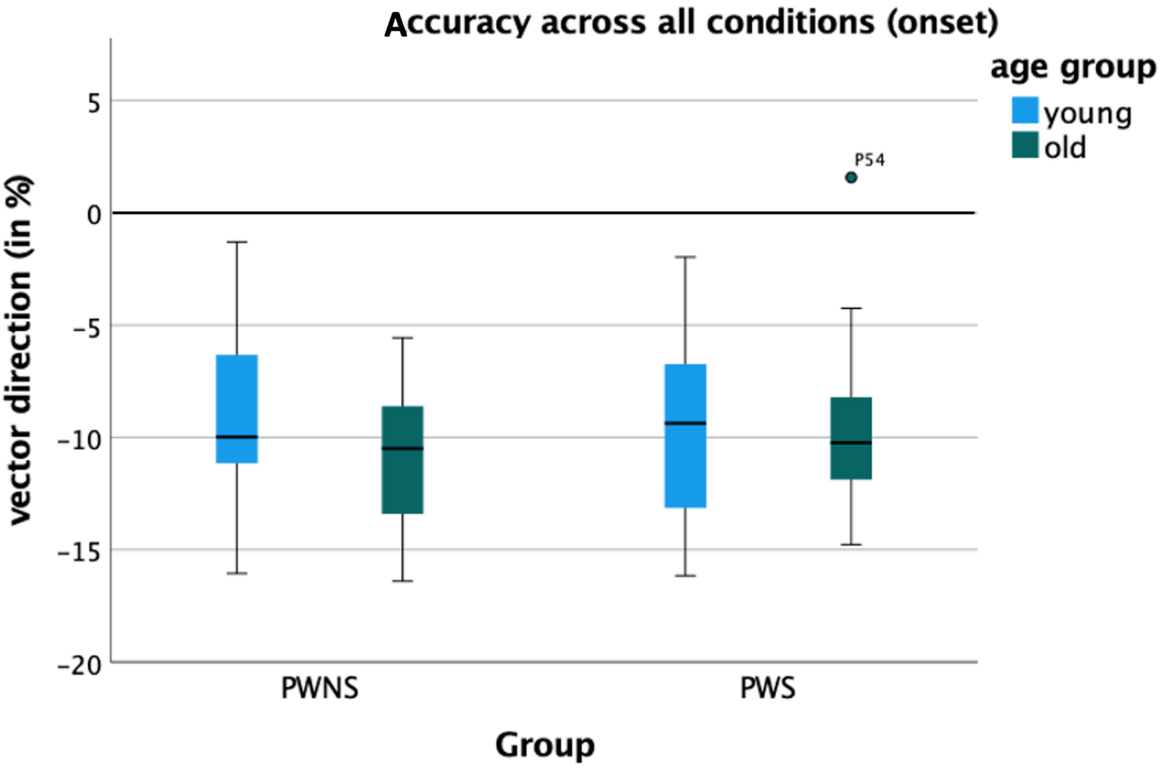


Figure 4.11 Accuracy (vector angle in % of the IOI) of synchronization across all conditions (onset as reference) in relation to the pacing event (=0).
Bars represent vector direction both groups subdivided for age groups and display the median; details as in figure 3.5.

To investigate synchronization accuracy in more detail for all different types of stimuli, figure 4.12 gives an overview of synchronization patterns for each condition and for the vowel as well as the onset in relation to the metronome. Here, the most important findings are summarized and visualized. This underlines what the ANCOVAS had already indicated: PWS are always later in their synchronization compared to PWNS concerning both reference points, the vowel, and the onset (see green line for a visual comparison of groups). It therefore does not seem to be a vowel-specific timing delay that is accountable for these results, but rather a general delay in word production. Furthermore, it becomes evident that with rising demands in articulatory complexity, both groups tend to start earlier (from latest start in simple syllables to earliest start in complex words). For the onset condition this effect of stimulus material was shown to be significant.

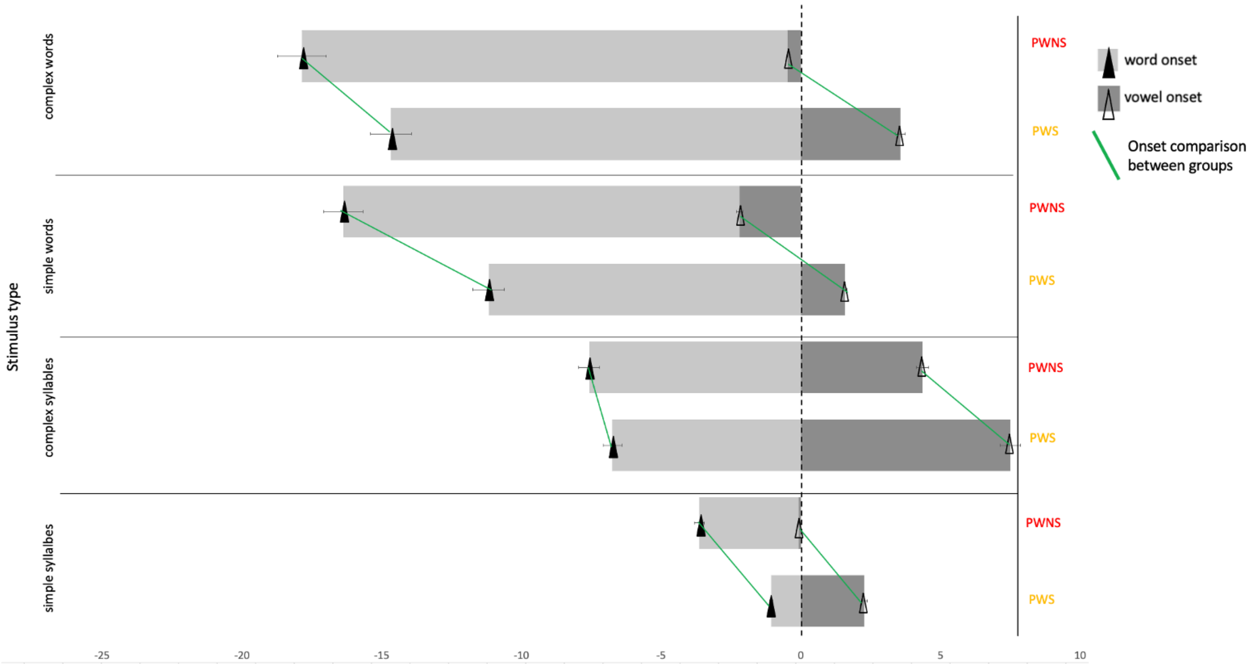


Figure 4.12 Mean accuracy (vector direction in % of the IOI) for the word onset and the vowel onset in relation to the pacing event (=0; metronome) for PWS and PWNS with age groups merged.
Error bars represent 95% confidence interval.

Analyses on accuracy were followed by analyzes on consistency. Whereas the first concept stands for the actual preciseness with which a participant is synchronizing to the metronome (how close to the pacing event is he/she with the vowel or onset he/she utters), the latter describes the variability, i.e. how evenly a participant synchronized. Therefore, only the vowel is investigated since this value rather stands for the reoccurring distance between uttered syllables/words.

Before analyzing data in detail, a visual overview of the four conditions shall be given in figure 4.13 for a first impression on consistency in PWNS and PWS as well as age groups: for both groups, consistency seems to be higher in syllables than in words. Furthermore, it seems that for syllables young participants seemed to display a more consistent performance in synchronization, whereas in the synchronization task with words older participants, especially within the group of PWS show a high consistency in their performance. Young PWS, in particular, seem to struggle with their consistency in synchronizing with words, though. However, performance regarding the consistency in synchronization seems to be comparable between groups as they are both ranging around a value of 0.8 and 0.9 for vector length. Detailed analyses shall clarify if there are systematic differences between groups or age groups with specific respect to different types of stimuli.

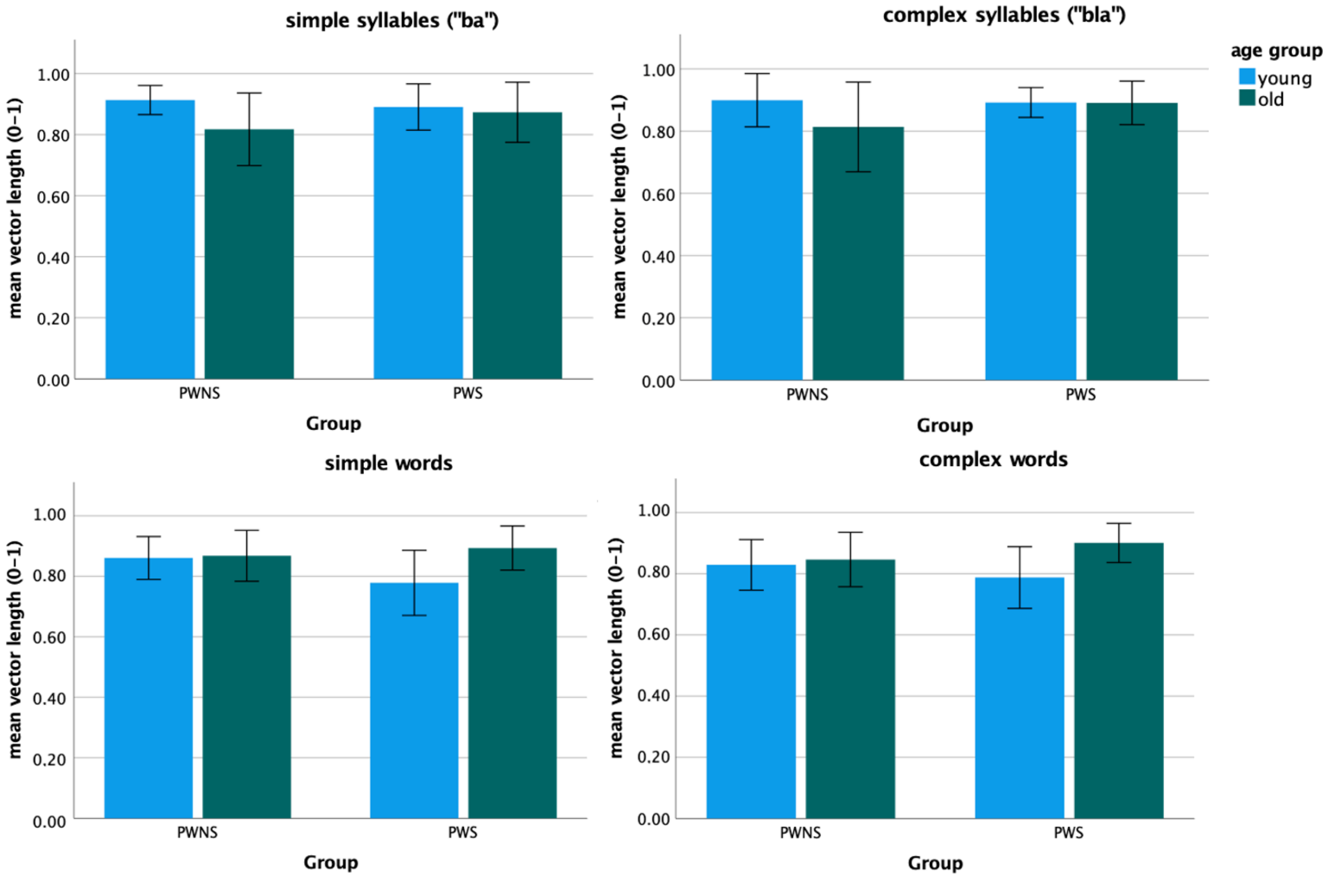


Figure 4.13 Mean consistency (vector length) of synchronization for each condition (vowel as reference).
Error bars represent 95% confidence interval.

Prior to the ANCOVA, consistency data were checked for normal distribution and homogeneity of variances: not all variables (simple syllables in PWNS, complex syllables in PWS, simple words in PWNS and complex words in PWNS) were normally distributed for PWS and PWNS; in young and old participants some variables lacked normal distribution, namely simple syllables in old participants, complex syllables in young participants, simple words in both age groups and complex words in young participants. The remaining variables showed normal distribution as assessed by the Shapiro-Wilk-Test; Sphericity was controlled for with the Mauchly-Test. Levene's test also indicated a given homogeneity of variances for simple and complex words, but not for the syllable condition. Values exceeding the third interquartile range were excluded from the analyses, since they indicated a rather random reaction instead of a real synchronization.

With regard to consistency (as measured by vector length), no main effects of group or age group were present. Instead, there was an interaction of stimulus material (syllable versus word) and age group ($F(1, 72) = 11.829$, $p < .001$., $\eta^2 = .141$). Decomposing this interaction no significant effects were present any longer; data did, however, show the trend that age groups nearly differed when reading words (*p* = .053), but not when repeating syllables (*p* = .68). As can be seen in figure 4.14, old participants were more consistent in synchronizing words to the metronome than young participants (mean vector length$_{old}$ for words = .890; *SD* =; .130 mean vector length$_{young}$ for words =. 820, *SD* = .130). These findings indicate a more stable timing performance in older participants when it comes to reading words, which might be related to more skilled reading ability in this age group.

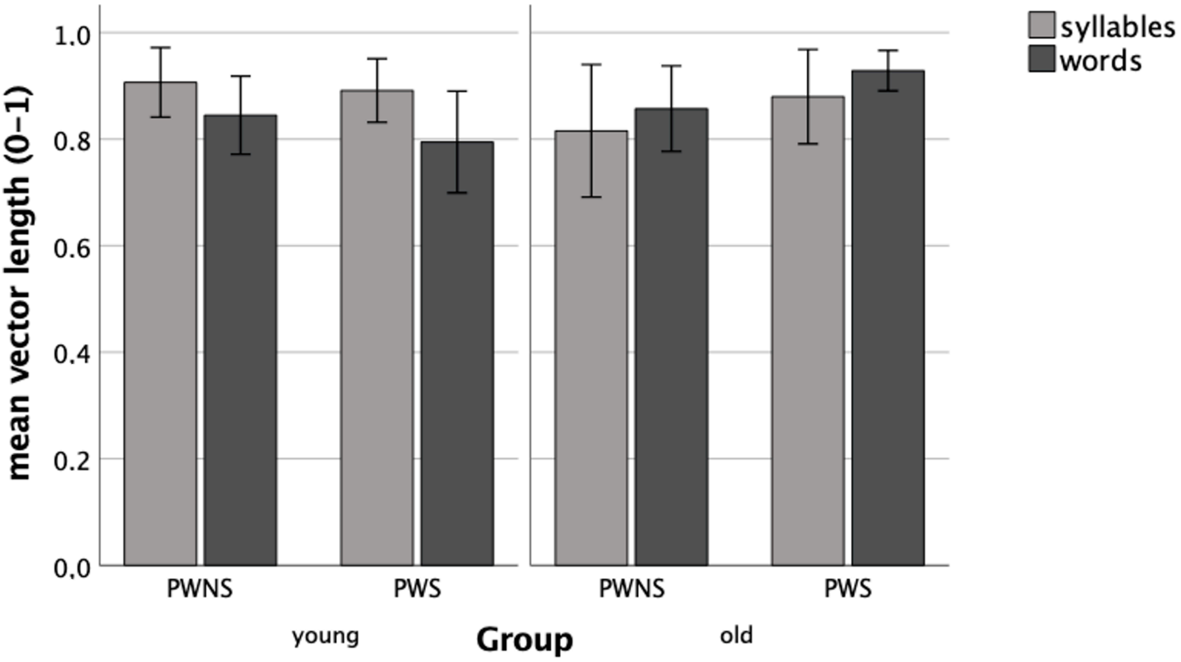


Figure 4.14 Mean consistency of paced speaking with a metronome (in vector length) in PWS and PWNS split by age group.
Error bars represent 95% confidence interval.

After comparing groups (PWNS versus PWS; young versus old) on measures of accuracy and consistency to carve out specific differences between those populations in their ability to verbally synchronize to a pacing event in different conditions, influential factors will be investigated in the following paragraph using correlations.

4.3.3 Influence of age and stuttering severity on synchronization performance

Significant results between groups have been found regarding accuracy for both variants of the synchronization task, namely vowel and onset accuracy. To further investigate these significant differences between PWNS and PWS, correlations (Spearman-Rho) with age and with stuttering severity will be conducted to see if they were an influencing factor here.

As the task was designed as reading task in the case of the simple and complex words, the hypotheses can be postulated as follows: old participants are expected to display a higher accuracy, since they are more trained in reading. However, neither for vowel nor for onset accuracy was a significant correlation with age found, indicating that this component of verbal synchronization does not develop within the age span investigated in this study (all $p > .05$). Calculating this correlation separately for mean word accuracy and leaving out accuracy on syllable synchronization, results display no significant effects either. Even when conducting analyses separately for each group (PWNS and PWS) no significant correlations were found. Hence, in PWNS and in PWS the ability to verbally synchronize to a pacing event was not influenced by the factor age, at all. This was true for the vowel synchronization as well as for the onset synchronization and different conditions such as the synchronization to words compared to syllables or the synchronization to simple versus complex stimuli.

A further relation that was under investigation in the context of significant differences found between PWNS and PWS was the influence of stuttering severity within PWS on their performance in verbal synchronization. It was hypothesized that PWS with more severe symptoms might also show less accurate performances. However, data showed that this hypothesis was not true since no significant correlations between initial stuttering severity and accuracy in vowel or onset synchronization was found. Even when subdividing conditions and investigating correlations separately (for simple versus complex stimuli and syllables versus words) all results remained below significance (all $p > .05$). Stuttering severity therefore did not seem to affect results on accuracy and unlike chapter 3 the removal of syllables/words affected by stuttering symptoms lead to a less direct impact on the fluent parts of speech. However, since significant differences between groups were found regarding accuracy, there seems to be a factor in speech planning or execution that is significantly affected by stuttering – it therefore seems to be linked to stuttered speech somehow, although it does not seem to be linked directly to stuttering severity.

All significant differences found on accuracy measures, therefore, are to be ascribed to differences in articulatory planning and execution in the population of young and adolescent PWS. The following discussion summarizes and interprets these results, recapitulating them in the context of current literature on this topic.

4.4 Discussion

In this chapter, verbal timing abilities in children and adolescents who stutter were investigated and compared to an age- and gender-matched control group. The focus of this research was to find patterns of predictive timing in those groups that may reveal systematic differences between groups of PWNS and PWS. To do so, a task of sensorimotor synchronization was chosen to compare various conditions in the verbal domain, such as unpaced and paced tasks, or syllable versus word synchronization as well as simple and complex stimuli.

Starting with the most interesting findings, results are in line with the first hypothesis of an altered predictive timing in terms of lower synchronization accuracy (Falk et al., 2015): despite the fluency inducing condition of an external pacemaker (metronome) PWS revealed a poorer performance compared to PWNS in terms of accuracy in the verbal synchronization task. PWS displayed a later vowel onset compared to the fluently speaking control group as they displayed larger positive lags between the pacing event and the vowel across all pacing conditions. PWS consistently timed their speech production later to the beat compared to the control group Therefore, it was further investigated if this was only the case for the vowel or if the onset of the syllable/word was affected by this delay in articulation as well. Results demonstrated clearly that this delay was not only observable for the vowel as reference but also for the onset, with data revealing a significantly later syllable/word onset in PWS. They therefore displayed a significant difference in the onset-vowel-timing to the beat. Hence, these results do support the idea of aberrations in articulatory movements of PWS, even when producing perceptually fluent speech (McClean et al., 1990; G. N. Zimmermann, 1980). A delay strategy rather than a problem within the transition from consonant to vowel seems reasonable here as an explanation, since both vowel and consonant show significant delays compared to PWNS. Also a reduced coupling of auditory and motor systems could be causal here, since reactions to the pacing event in PWS differed significantly to PWNS (Civier et al., 2010; Harrington, 1988; van de Vorst & Gracco, 2017).

An over-anticipation, as observed in other studies investigating SMS in PWS (Falk et al., 2015; Olander et al., 2010; Sares et al., 2019) cannot be confirmed when using the vowel as reference. The vowel, as the center of the syllable/word was shown to have positive lags in relation to the pacing event, therefore, the phenomenon of NMA (see chapter 2.6.3.1) cannot be reported for the vowel. However, when choosing the onset of the syllable/word as reference, a NMA is observable in both groups, stronger though, within the group of PWNS. This tendency to anticipate a pacing stimulus is something very common and frequently reported in research, often under the name of NMA: negative mean asynchrony stands for the observed phenomenon that asynchronies, i.e. synchronization errors tend to occur some milliseconds prior to the actual pacing event and are therefore not distributed symmetrically around it (Aschersleben, 2002; Falk et al., 2015; Repp, 2005; Repp & Su, 2013). The NMA can bring relevant information into the question about underlying mechanisms in deficient timing systems. However, this study reports different results compared to Falk and colleagues (2015), since they found the tendency to anticipate to be bigger in stuttering than in non-stuttering children and adolescents. It is a fact though, that NMA tends to get smaller in musically trained persons which is a factor that must be kept in mind when interpreting these results, since musical training was significantly higher within PWS compared to PWNS. A further fact that cannot be confirmed for this data, though, is that NMA tends to get smaller with more complex stimuli, such as in our case – the wordlists which demanded a higher tempo in synchronization than syllables and especially in complex words which demand higher articulatory capabilities. In case of the data at hand, NMA got bigger with complex words compared to simple words and compared to syllables. The consistent bias towards over-anticipation in stuttering participants could not be demonstrated here, since only in relation to the pacing event and not in relation to fluently speaking participants and only in case of the onset, but not in case of the vowel, was over-anticipation found. Summing up the most important findings first, all results will be discussed in detail, following the chronology of analyzes.

For the unpaced condition no differences between groups could be discovered, indicating the timing deficit is specifically prominent in tasks of sensorimotor synchronization that require an integration and coupling of auditory-motor abilities. The investigation of mean IVIs to compare the average chosen tempo when uttering syllables/words as well as the investigation of the CV of the IVIs to compare the variability in this task between PWS and PWNS did not show any significant differences. This result was also found for age groups. Even though lacking significance in those group and age group differences, graphs showed some tendencies: firstly, it hinted towards the fact that PWS, especially young PWS, show a higher mean IVI than PWNS. Secondly, for all groups besides old PWS, the mean IVI was higher in syllables than in words, which seems a little surprising, since syllables are shorter and easier to articulate than words which would rather lead a shorter interval to be expected. The third observation of figure 4.3 is that the mean spontaneous IVI for syllables and for words lies between 800 to 1000ms. Considering the tempi chosen for the synchronization task, this interval is closer to the one chosen for the synchronization to words (900ms), whereas the one for syllables (750ms) was faster than any tempo chosen by participants themselves. Maybe this could hint towards the fact that the pace chosen for the syllable synchronization was too fast. Looking at articulatory variability in the unpaced task, results revealed a higher variability in PWS when reading words, but very similar results between age groups and groups when uttering syllables. However, all results for the comparison of the CV of the mean IVI stayed below significance, indicating a comparable performance between groups with only tendencies revealed by figure 4.5. A significant effect of stimulus material was found, hinting towards the fact that more complex stimuli (i.e. words) lead to a higher variability as opposed to syllables. An interaction of stimulus material and age group revealed the fact that there does not seem to be a big difference between age groups when looking at syllables, words however showed a higher variability in young participants as opposed to old ones. Results, however, stayed below significance, once resolved with t-test and can therefore only be interpreted as trends. Still, this might hint towards the fact that, especially within younger participants, complex stimuli such as words also lead to a higher variability. One fact that has to be kept in mind when interpreting results of the unpaced condition is the significant difference between PWS and PWNS regarding the duration times: not only the mean vowel duration, but also the mean word duration was significantly longer in PWS compared to PWNS. This imbalance was accounted for by adding an average duration of all conditions as a covariate in the analyses. However, when interpreting graphs and looking at tendencies it must be considered that PWS already show differences in the unpaced condition, when it comes to the mere duration of vowels or words within this task. Although no significant differences between groups were found for the unpaced tasks, it is an important insight into the verbal production of PWS. Longer production times alone hint towards the fact that there are processes during the perceptually fluent articulation of PWS that differ from those of PWNS.

Investigating the paced tasks, the question about the variability-reducing effect of the pacing event was to be answered first: as pacing is known for the stabilizing effect on motor variabilities (Andrews et al., 1982) differences between the CV of the IVIs for the unpaced and paced condition were investigated. It became clear that both groups benefited from the pacing event in terms of lower variability, but the percentage of participants displaying a reduced variability was higher amongst PWNS compared to PWS. Although results on group comparisons regarding the changes in variability induced by the metronome stayed below significance, percentages and figure 4.6 revealed the tendency to benefit more from the pacing event in PWNS compared to PWS. This finding could hint towards a speech motor production system that is characterized by more unreliable timing mechanisms; they actually benefit from an external pacemaker but still cannot be compared to those of fluently speaking persons, as they do not respond to pacing as strongly as unimpaired systems in PWNS (Civier et al., 2013).

Analyzing data on accuracy (for the vowel) and interpreting the results, it becomes obvious that across all conditions, PWS produced the vowel onset significantly later compared to PWNS. This finding already supports the idea of a compromised speech system struggling with complex demands such as the paradigm of a verbal synchronization task. (Usler & Walsh, 2018). However, a comprising interpretation will follow after the recapitulation of all results on paced performance.

Results for data on accuracy concerning the onset as reference repeated the significant differences between groups with PWS starting significantly later compared to PWNS. Analyses on the onset also revealed an effect of stimulus material: in both groups, syllables started later compared to words, which is visualized in figure 4.10. However, accuracy data did not show any effects of age group, implying that within the age span tested no significant development/improvement in articulatory processes or synchronization ability takes place.

Continuing with results on consistency, they revealed no group differences. PWS and PWNS therefore seemed to be evenly consistent in their synchronization but with different distances to the actual pacing event, as differences in accuracy revealed. Despite no group differences being present for consistency, an interaction of stimulus material and age group revealed that age groups performed differently when reading words but not when repeating syllables. After re-analyzing the significant interaction with a robust test, significance was not present anymore, but trends were still visible: old participants were more consistent in synchronizing to words than young participants – this effect was not observable for syllables, however. An explanation for this result could be the more mature reading skills in old participants.

Regarding the correlations conducted, no improvement of synchronization ability was found with rising age and furthermore, no link between more severe stuttering and poorer synchronization abilities. These results are comparable to a study by Hilger et al. (2016) who could not demonstrate a relationship between the performance on the motor timing task and stuttering severity or the likelihood for recovery from or the persistency of stuttering. Note, however, that the task under investigation differed, since they investigated a bimanual hand clapping task. As any further correlations failed to reach significance for the influence of age or stuttering severity on the observed results, the hypotheses postulated at the beginning of this chapter can be answered as follows: speech production to an external pacemaker - although it might lead to more fluent speech – still shows signs of altered predictive timing in terms of lower synchronization accuracy (Falk et al., 2015). However, there was no difference in terms of accuracy or consistency between young and old participants. Stimulus material and stimulus complexity had a significant influence in some cases – as expected more complex stimuli (words or complex onsets) also lead to a later production in PWS. However, this effect was not observable for all conditions under investigation.

Still, the question about the origin of those deficits in accuracy must be solved. They can derive from various dependent or independent sources, that need to be evaluated and linked to speech timing and stuttering. These differences in synchronization performance, that can be described as a timing delay, indicate a specific timing deficit which could be a result of at least two temporal processes: On the one hand, altered temporal predictions in individuals who stutter may lead to delayed temporal targets during production (Harrington, 1988), on the other hand it could also be more unreliable timing mechanisms which may generate delays in the activation of syllable motor programs during articulation in PWS (Civier et al., 2013). A combination of both processes might be possible as well. Altogether, these recent findings support the idea of altered timing in young and adolescent speakers who stutter. Unfortunately, the results cannot answer the question if the found differences in PWS stem from motor or predictive timing origin. This question must be answered in future research and leads to limitations of the study at hand: a precise differentiation between the two potential causes is not possible, since the testing paradigm does not allow to test separately for either the motor or the predictive timing component. Since the task chosen involves both the ability to predict the pacing event and one's own verbal response to it as well as the motor component when reacting verbally to the pacing event, there is no way to investigate both processes taking place in a separate manner. A testing paradigm involving motor responses in sensorimotor synchronization always relies on the preceded steps of perception and prediction (of pacing events and motor responses) and is therefore not suited to answer this question – other than for example a study from Wieland et al. (2015) which focused on the perception only and found evidence for a deficit in children who stutter. Testing the perception exclusively is hence possible, testing the motor component without the preceded part of the perception unfortunately not, since the basis to every motor reaction in form of a synchronization is the process of perception.

A further limitation of the study conducted is the limited number of participants, especially when subdividing them into age groups numbers get quite small for generalized statements and the transfer of results. Despite the fact that the number of 40 participants in the group of PWS and PWNS is actually quite comparable to many peer- reviewed and internationally published articles within stuttering research in this age group (see Falk et al., 2015; Franke et al., 2021; Wieland et al., 2015), the number of participants is reduced for various tasks due to outliers that needed to be removed leading to a reduction in total participant number. Furthermore, the number of participants within the age groups is not balanced perfectly, as explained before, with more young than old participants.

Another limitation that needs to be discussed here is the fact that all calculations had to be conducted with a covariate: mean syllable/word duration. Musical training differed significantly between groups, as well. However, as correlations with any of the parameters of SMS tested did not show any significant coherence and therefore, a direct influence on synchronization performance is not given. Despite the knowledge that verbal synchronization is very similar to abilities improved in musical training such as the perception of and the response to given rhythms (Ladány et al., 2020; Repp, 2010; Repp & Su, 2013) it was not added as a covariate to calculations, since analyses proved there was no correlation. Mean duration times, however, which differed significantly between groups have direct impact on synchronization, as the mere duration is crucial to the way a verbal synchronization takes place. Therefore, the decision to incorporate mean duration times into the analyses is logical as well as reasonable, which lead to the decision to conduct analyses as ANCOVAs without overloading the complex model.

In sum, the thesis at hand faces limitations that must be considered when interpreting results; however, those limitations are quite common and sometimes inevitable when conducting studies with such populations.

Relating the findings of this study to existing results and neurological findings on stuttering in children and adolescents it becomes evident that it is not surprising to find even the perceptually fluent speech of PWS to be aberrant compared to PWNS: even in young persons affected by stuttering neuroanatomical and neurofunctional aberrations were found, mostly within the BG (Beal et al., 2013; Chang & Zhu, 2013) but also in further areas belonging to the timing network (Foundas et al., 2013). Those results hinting towards emerging compensation within the brain of young PWS can therefore explain why perceptually fluent speech still is not just the same as fluent speech of PWNS – even under fluency enhancing conditions. The fact that even at such a young age mostly permanent changes to the brain have taken place is most likely to be measurable within output produced by the brain – in this case: speech. Aberrations within the connectivity, structure or function of exactly those brain structures that are also relevant for processing and producing rhythm or speech do not vanish completely even if speech motor output is fluent and therefore sounds “normal” at first sight. Articulatory processes are combined of so many fine-tuned steps that an apparently unimpaired verbal output might still show various signs of impairment when having a closer look. As in chapter 3, articulation rate differed significantly between PWS and PWNS despite the removal of all instances of disfluencies. This chapter aimed to take an even closer look at evidently fluent articulation in PWS by analyzing onset and duration times as well as measures of synchronization, and comparing them between groups. It therefore seems only reasonable that – when looking at verbal output in more detail – aberrations found at a larger scale (such as rates) are measurable on the level of onsets or vowels in syllables/words. Despite the analyses conducted in this chapter being more "detailed" compared to the previous chapter, there are many ways of analyzing speech of PWS in even more detailed and sensitive ways, such as kinematic analyses (McClean et al., 1990; Smith et al., 2012; Wiltshire, Chiew, Chesters, Healy, & Watkins, 2021) that might reveal different results due to the fact that kinematic and acoustic analyses are not comparable with each other.

These results do not only provide evidence for a rhythmic deficit in children and adolescents who stutter, they also provide the chance to use this knowledge for an additional purpose than only the mere investigation of differences: Summing up the results of all the studies cited within this thesis and adding the most recent information of the results just discussed in this very chapter, it becomes clear that rhythm does not only play a crucial role within speech perception and production in general but it most certainly shows aberrations within the population of PWS. Those aberrations were evident in different modalities, such as speech and non-speech movements as well as in different contexts (musical rhythm versus speech rhythm versus mere metronome synchronization) and in both domains – perception and production. Those behavioral results are supported by results of neurological and neurofunctional evidence in PWS during rhythmic tasks. When adding up those results one might ask why this vast amount of information on this specific topic is until now mostly used for investigative purposes (causes of stuttering) but not for further purposes such as the diagnosis or the therapy of stuttering. Of course, it is very reasonable to investigate causes of stuttering – as previous chapters and especially chapter 2.5 have shown, there is still a lot to be discovered and questions to be solved. However, if this information is collected it might as well be used for further purposes, such as additional diagnostics or therapeutic intervention in stuttering. Therefore, the following chapter aims at doing this exact thing: Using the rhythmical information collected in PWS to turn it into a tool that might be helpful for therapeutic intervention.

5. Predicting stuttering therapy outcome with temporal parameters

The “Atypical Rhythm Risk Hypothesis” (Ladány et al., 2020, p. 1) postulates that persons with “[…] atypical rhythm processing are at higher risk for developmental speech/language disorders .” (Ladány et al., 2020, p. 15). Stuttering might be one of those disorders, as the previous chapters have shown. Considering the questions that are of utmost importance to nearly all parents of CWS, rhythm might be a powerful and sensitive tool to answer those questions more precisely and more satisfactorily than so far: most parents are concerned with the question whether stuttering will persist in their child – a question that is impossible to answer distinctively yet. Furthermore, they are interested in therapies and the potential outcome for their child, which also is a question that cannot be answered yet. At this point, temporal parameters and rhythm in its different variations bring in a powerful potential to shed more light on those important questions raised. Of course, it must not be mistaken for a diagnostic tool giving definite statements about developments that cannot be foreseen thoroughly. However, (impaired) timing skills and the screening of those can give valuable information about risk factors for persisting stuttering and the potential success of therapeutic interventions at low effort and costs (Ladány et al., 2020). The following chapter aims to investigate the power of temporal skills and rhythmical screenings as tool to predict therapeutic success in a population of children and adolescents who stutter.

5.1 Introduction

The start of a therapy is often accompanied by feelings of hope, enthusiasm, and excitement – not only in PWS but also in their close relatives or, in the case of CWS, their parents. Expectations are high, searching for a cure from a speech disorder that can impact daily life enormously (Guitar, 2014; Starkweather, 1980). Depending on the expectations prior to therapy, results of the intervention can leave PWS and their close relatives disillusioned to various reasons: sometimes, expectations simply are too high, and a complete recovery of stuttering can only be accomplished in some cases, strongly depending on age. Furthermore, therapeutic interventions differ a lot and not every therapy is suited for any PWS. Finally, the success of a therapy also depends on the commitment and effort of the affected person and the support of persons surrounding (Guitar, 2014; Thum & Mayer, 2014). However, neither does this imply that only a complete recovery from stuttering is a success nor is it the fault of PWS if therapeutic interventions do not show the expected results. These are only a few of the external factors influencing the result of a therapy – besides many others. Finding out which factors are relevant for the further path – persistent versus recovered stuttering or a good therapy outcome versus non-respondence to the intervention – is still a question to be solved.

Research on the factors influencing persistency versus recovery in stuttering has been trying for many years, even decades, to figure out which factors are crucial for the further development of stuttering within a child affected by this disorder. As explained in chapter 2.3.5 some children just stop stuttering with the help of therapy or even without intervention and some persist to stutter – despite intensive therapy. Finding the factors that make the difference for the further path of a child affected by stuttering is highly relevant. Besides the known factors such as age (at onset), gender and family history of stuttering, a more specific screening tool could provide valuable information for professionals and parents (Leech, Bernstein Ratner, Brown, & Weber, 2017; Yairi & Ambrose, 1999). Many authors have tried to develop models; for example like Howell and Davis (2011a) achieved nearly 80% specificity and sensitivity in predicting whether eight-year old CWS will recover or persist by teenage years. Other authors have tried to identify differences on neurological levels (Chang et al., 2008), in motor abilities (Ambrose et al., 2015), or in language skills (Leech et al., 2017). Also, the influence of speech and articulation rate on the further path of stuttering within CWS was investigated, displaying a tendency for children who later recovered from stuttering to show slower rates than persisting children (Hall et al., 1999). It is mostly tendencies that can be extracted within these studies. Hence, research still needs to provide more robust results.

Research on therapy outcome is a topic of high interest, as well, since not only PWS, but also their parents or close relatives are interested in the potential outcome of a therapeutic intervention. Therapy demands lots of time, effort, money, and engagement of the person affected and their therapist and leads to high costs for the health care system, as well. It, therefore, is not only a question of personal interest for the person affected, but rather a decision how the current available interventions and capacities can be used wisely, and maybe even which therapeutic intervention is most promising for whom.

The core component and aim of the everyday life of many who stutter is the reduction of stuttering symptoms in speaking situations. Many PWS who have undergone therapy struggle to maintain the benefits of learned speech-restructuring treatment, with only one third managing to maintain learned fluency (Craig & Hancock, 1995). Therefore, it seems reasonable to look for factors that might predict the outcome of a specific stuttering therapy. General factors like pre-treatment stuttering severity, personality and attitudes about speaking were investigated regarding their influence on the outcome (Guitar, 1976). But also more specific factors like co-occurring mental health disorders, psychosocial impact, parental personality, language skills and genetics have been taken into account when investigating factors which influence therapeutic success (Andrews & Craig, 1988; Frigerio-Domingues et al., 2019; Iverach et al., 2009).

Two recent studies that were conducted with CWS and are consequently very relevant for the thesis at hand were implemented by Cook and colleagues (2013) and Park and colleagues (2021). Whereas the first assessed 3 factors only concerning CWS, namely stuttering severity, lexical diversity, and psychosocial impact to predict outcome after an intensive therapy course, the latter investigated 10 variables aggregated within the 5 categories of demographics, parent stuttering severity measures, child speech and language measures, child psychology measures and parent psychology measures. Cook and Howell (2013) investigated stuttering severity and psychosocial impact (Cook, 2013) prior to therapy – two diagnostics that are conducted prior to therapy anyway as well as lexical diversity as measured by Type Token Ratio in fifty-four CWS. This way of collecting information prior to therapy seems very efficient, since only the collection of Type Token Ratio is an additional measure. This ratio is the result of the division of the total number of unique words (i.e. types) by the number of all words uttered within a given excerpt of spoken language (i.e. tokens) and hence is a very efficient way of investigating lexical diversity (Cook et al., 2013). Assessing therapy outcome by linear and logistic regression analyses, only initial stuttering severity was a significant predictor for therapy outcome as measured in stuttering severity. Besides regression analyses, correlations were conducted, indicating a correlation of psychosocial impact with improvement in fluency as well as a correlation of lexical diversity and therapy outcome. Unfortunately, this is not a very specific result enabling therapists to give precise prognostic statements (Cook et al., 2013), however it is in line with previous results (Howell & Davis, 2011a; Starkweather & Gottwald, 1993). The study conducted by Park and colleagues (2021) is very comprehensive, using a total of 32 variables as potential predictors for therapy outcome in the Lidcombe program. A cohort of 277 children and their parents were included in the study. Since the Lidcombe program was developed for early stuttering, it was only very young CWS (3 – 6 years) that participated in the study. As mentioned before, the 32 variables were aggregated within 5 categories: Demographics contained typical information on age, gender, family history etc., parents rated their child’s stuttering severity on a scale, child speech and language measures were conducted with various clinical evaluations on expressive and receptive language, child and parent psychology measures were also assessed with approved clinical instruments. Using regression analyses, results showed that children with better language skills and a rather easy temperament showed better outcome. However, it has to be noted that these variables could only account for a small part of the variance of treatment outcome – variables that were identified as outcome predictors were only statistically, but not clinically significant (Park et al., 2021).

Former studies and their results reveal the importance of further investigation of outcome predictors, since they add value to children, parents, and therapists in the practical and clinical setting. Those predictors, however, must also meet some criteria to be applicable within the clinical setting: predictors for therapy outcome have to be specific, of course, but they also have to be feasible, meaning they must not be too time-consuming or complex, since diagnostic processes prior to therapy are mostly quite encompassing themselves (Guitar, 2014; Thum & Mayer, 2014). It therefore is a further goal of the thesis at hand to investigate whether articulation rate as very general and basic measure of speech rhythm as well as tasks of speech synchronization as rather specific rhythmical abilities, have the power to predict therapy outcome. It has been hypothesized before in the Atypical Rhythm Risk Hypothesis (Ladány et al., 2020) that atypical rhythmic skills in children might be a risk factor for disorders in speech and language; it also has been demonstrated within various studies that CWS show atypical rhythm in terms of auditory rhythm discrimination (Chang et al., 2016; Wieland et al., 2015), finger tapping (Falk, Müller, & Dalla Bella, 2015), metronome clapping (Olander, Smith, & Zelaznik, 2010), breath pauses during reading aloud (Franke et al., 2021) or verbal tasks such as speaking (M. Adams & Hayden, 1976; M. R. Adams & Ramig, 1980; Andrews et al., 1982; Hulstijn et al., 1992; Sares et al., 2018) or singing (Falk et al., 2016). Recapitulating these results, Ladány and colleagues (2019) envisioned a risk factor model using rhythmic tasks to screen for signs of atypical rhythm that might be used as predictor for the risk of developing stuttering (as well as other speech and language disorders). However, rhythm could not only be used as a kind of screening tool for later development of speech and language disorders, including stuttering, but it can also be hypothesized that rhythmic tasks might be a useful tool for the prediction of therapy outcome.

In analogy with the normalization of white matter and activation patterns after successful therapy or recovery from stuttering it is a logical consequence that with the normalization of speech fluency and related neural correlates also (speech) rhythm and its neural correlates approach rhythmical skills of fluently speaking persons. It therefore might seem reasonable that PWS with rhythmical skills that only show minor aberrations or in other words – who show rhythmical skills that are more like those of PWNS than of PWS – tend to have better outcomes than those with strong aberrations within their rhythmical abilities and therefore also on neurological levels (Kell, Neumann, Behrens, von Gudenberg, & Giraud, 2018; Kell et al., 2009; Ladány et al., 2020). Given the fact that results on parameters like language skills, child and parental personality, or genetics haven`t reached statistical significance or clinical evidence, and, at the same time those parameters were effortful and complex to collect, it seems reasonable to try out new paths within the research on prognostic factors on therapy outcome in stuttering. So far, no study has investigated this possibility yet. Combining the results from studies on rhythmical aberrations in PWS with the investigations on therapy outcome predictors, it might be worth trying to add rhythmical abilities as variables in regression analyses. Thereby, new additions to already existing efficient variables, like stuttering severity, could emerge. The investigation of complex and effortfully collected variables derived from the language of CWS or psychological parameters of the child or the parents proved to be inauspicious, therefore new ways are required (Cook et al., 2013; Park et al., 2021; Reed & Wu, 2013).

According to the current state in literature, hypotheses are postulated as follows: Participants with high initial stuttering severity as measured with SSI-3 will be more likely to persist and participants with mean articulation rates that are closer to those of fluently speaking ones are expected to show a better outcome of therapy. Furthermore, participants displaying rather accurate verbal timing skills are more likely to have a better outcome, as well.

The practical relevance of this topic and the clinical utility is clear: Predicting from pretreatment characteristics which person is presumably going to benefit from the intervention, enables therapists, PWS themselves and their surroundings to also take remedial action prior to a possible relapse. On top of that, knowledge about predictors of stuttering intervention outcome enhances practice on an evidence-based level. Prognostic statements to PWS and their parents or relatives could be a lot more precise if treatment outcome could be predicted by information from case history and patient variables (Andrews & Craig, 1988; Park et al., 2021). A first attempt to use temporal parameters as a screening tool is tested in this chapter.

5.2 Material and methods

5.2.1 Participants

54 native German-speaking children and adolescents who stutter participated in the experiment. Due to comorbidities (i.e., ADHD, dyslexia, cluttering) or cognitive impairment, 15 participants had to be excluded from the analyses. Since the comorbidities of cluttering would influence

performance on articulation rate, and dyslexia and ADHD could falsify results on reading and SMS, participants affected by these disorders had to be excluded (Canhetti de Oliveira et al., 2013; Huss et al., 2011; Noreika et al., 2013; Thomson & Goswami, 2008). The final sample therefore consisted of 39 participants (4 females, 35 males) with developmental stuttering (mean age = 12,8 years; SD = 2,3 years).

Participants who stutter are in the following chapter abbreviated as PWS, to avoid confusion, since CWS would not cover the age span of all participants and lead to confusion when talking about age groups. PWS were recruited with the help of a therapy course held near Munich during the summer of 2017, 2018, 2019 and 2020 (staerker-als-stottern.de). The concept behind the therapy course can be described as a method combination of fluency shaping and modification techniques (see chapter 2.4.4.5), with additional psychological coaching and relaxation techniques. Such a concept aims at combining the advantages of both, fluency shaping and modification (for mor information see chapter 2.4.4.4). It is the goal of method combination to enable patients to choose for each situation which techniques they want to use and which grade of fluency they aim to achieve. For the collection of the data and hand, participants were asked to use a technique that helps them to reach a high level of fluency The first collection on data about stuttering severity was, of course, done prior to therapy to avoid effects of therapeutic intervention.

Stuttering severity was assessed with the SSI-3 (Riley, 1994) and the German Version of the OASES (Yaruss & Quesal, 2006). Whereas the SSI-3 scores quantify objective measures of stuttering severity, the OASES is an assessment of the subjective psychosocial impact of stuttering on the everyday life of the participants. More details about the diagnostic process are explained in the chapter to follow. The SSI-3 scores ranged from mild to very severe stuttering with a mean of $M = 26.97$ ($SD = 10.86$), the scores for the psychosocial impact from very mild to severe in the initial testing session with a mean of $M = 2.38$, $SD = 0.52$. All the tests related to rates and rhythmical abilites were conducted prior to therapy, as well, but in a separate testing session scheduled for the next day. Finally, stuttering severity was measured again, after completing the intensive therapy course, which lasted for 15 days, and the same instruments were used (SSI-3 and OASES). Within this session, SSI-3 scores ranged from very mild to severe stuttering with a mean of $M = 14.08$ ($SD = 8.82$), the scores for the psychosocial impact from mild to moderate with $M = 1.96$ ($SD = 0.4$). The exact distribution of severity categories for SSI-3 and OASES pre- and post-therapy are summarized in table 5.1. As the table already displays, there has been a clear improvement regarding the subjective and objective stuttering severity. A more precise analysis of the improvement during the intensive therapy course will be conducted in chapter 5.3.1.

In addition, age groups were formed to allow for analyses between young (9-13.25 years, "young") and old (13.75 -17 years, "old") participants. There were 22 (3 female, 19 male) PWS in the young group and 17 (1 female, 16 male) in the old group. To compare the results for both age groups an independent samples t-test was calculated. Data on stuttering severity pre- and post- therapy and on the subjective influence of stuttering showed homogenous distribution of variances and no significant differences between age groups. Data on mean articulation rate and mean accuracy showed homogenous distribution of variances as well and no significant differences (all p > .05) between age groups. Hence, no significant differences between the two age groups were found regarding any of the variables tested and relevant for the upcoming regressions, so the two age groups can be merged or analyzed separately, depending on the questions raised.

Participants and their parents were informed prior to the study and gave informed consent. The study was, of course, approved for conformity with ethical standards by the ethics committee of the faculty of medicine of the Ludwig-Maximilians-University Munich.

Severity	**SSI-3 pre**		**SSI-3 post**		**OASES pre**		**OASES post**	
category	**n**	**%**	**n**	**%**	**n**	**%**	**n**	**%**
1	2	5.1	17	43.6	1	2.6	2	5.1
2	8	20.5	10	25.6	15	38.5	26	66.7
3	10	25.6	8	20.5	19	48.7	11	28.2
4	10	25.6	4	10.3	4	10.3	0	0
5	9	23.1	0	0	0	0	0	0

Table 5.1 Distribution of participants per severity category for SSI-3 and OASES (*n* = 39)
SSI-Categories: 1 = very mild; 2 = mild; 3 = moderate, 4 = severe; 5 = very severe;
OASES-Categories: 1 = mild; 2 = mild-moderate; 3 = moderate; 4 = moderate-severe; 5 = severe

5.2.2 Instruments and procedure

The goal of this study was to find out if parameters tested during the previous chapters (Chapter 3 and 4) can predict therapy outcome. The detailed implementation of stimuli and the step-by-step procedure can therefore be extracted from the chapters 3 and 4. For a comprehensive understanding, however, the most important steps are described as follows:

Prior to any rhythmical testing sessions, the assessment of stuttering severity was scheduled for day one of the intensive therapy course. The chosen instrument for the assessment of stuttering severity was the SSI-3 (Riley, 1994) to guarantee an objective quantification of stuttering symptoms. This assessment was done by speech therapists trained and specialized in stuttering who were working for the course. The SSI-3 consists of four parts: whereas the first part is on the quantification of stuttering symptoms as measured by the %SS during a spontaneous speech sample, and in the case of school-aged children an additional short text. Points are assigned progressively for the %SS. The second part is concerned with the mean duration of the three longest lasting symptoms; points are assigned depending on the duration – the longer the mean duration of the symptoms, the higher the number. The third part is about additional motor behavior (i.e. sounds, grimacing, head, or limb movement): The more perceptible and salient those motor behaviors are, the higher they are rated on a scale from 1-5. Afterwards, the sum of those ratings is calculated. The fourth part of the SSI-3 sums up all points from parts one to three and those raw values are transformed with normalized and standardized tables into grades of severity reaching from very mild to very severe.

The instrument for the evaluation of the subjective impact of stuttering on one's life was the German Version of the OASES (Yaruss & Quesal, 2006). The OASES is a self-reported measure to examine the entirety of stuttering from the perspective of persons affected by the disorder (Yaruss & Quesal, 2008). It is subdivided into four sections, concerned with general information on stuttering experience, one's reactions to stuttering, communication in daily life and the quality of life. Each statement or question must be answered by the person affected by choosing the grade of agreement on a scale encompassing 5 stages. Afterwards points of all items answered are added and impact scores are calculated reaching from mild to severe impact of stuttering on one's life (see also table 5.1 for the distribution of severity categories in SSI-3 and OASES).

The "instrument" to measure articulation rate was a little more complex: For collecting speech samples and speech synchronization results of participating children and adolescents they were submitted to an interview and reading session, lasting about 10 minutes, depending on reading abilites and severity of stuttering symptoms. Participants were asked to sit comfortably at a table where they were recorded with a ZOOM H4N recorder (44.1kHz, 16 bit). An external headset microphone (beyerdynamic opus 54.16/3) was used for optimal acoustic quality, recordings were done within a quiet room with the experimenter present during the whole session. For the interview, participants were asked comparable questions about their daily life and hobbies. The interview was supposed to last about 3 minutes at least to ensure the speech sample was sufficient and contained at least 60 seconds recordings of the participant talking. Afterwards participants were asked to read an excerpt from a German children's book aloud, that is popular within the age range tested and recommended for readers from 8 years on (Maar, 2008). Note that there were 11 participants within the group of PWS that could not attend this testing session. In this case, speech samples of the diagnostic session also consisting of an informal conversation about daily life and a read excerpt consisting of 250 syllables (on the topic of holidays) was used. The detailed data processing procedure is described in chapter 3.2. After raw data had been edited, articulation rate was calculated as follows:

$$Articulation\ rate = \frac{number\ of\ fluent\ syllables}{total\ duration\ of\ excerpt - duration\ of\ disfluencies\ (syllables + pauses) - pauses > 250ms}$$

Afterwards, articulation rates of speaking and reading were used to calculate a mean articulation rate that is being used for the statistics to follow. Articulation rate was chosen instead of speech rate, as it differed significantly between PWNS and PWS (see chapter3). On top of that – and besides the fact that speech rate also differed significantly – it is the more interesting and sensitive measure to use, as it is obvious that speech rate differs due to all disfluencies included - whereas in articulation rate all kinds of disfluencies had been removed.

The instrument to measure speech synchronization was quite complex, as well: for the task on speech synchronization, additionally to the technical set-up for the recording of articulation rate, Neusonic NE03 studio monitors were installed in front of the participant to present the metronome in the free field at a comfortable sound pressure level. Participants were asked to utter four different types of stimuli: simple, repeated syllables ("ba"), complex syllables ("bla"), words with simple onsets (e.g., "Lauch") and words with complex onsets (e.g., "Schleim"). The word lists contained 24 monosyllabic words each (12 nouns and 12 adjectives, see appendix 2). Each list was preceded by 5 more items to familiarize with the task. Those stimuli were chosen to control for syllable complexity as it might provide a better insight into ongoing processes of articulation in stuttering. A first and unpaced trial, where participants were asked to repeat the syllables and to read the words at a self-chosen comfortable pace, but with the instruction to speak as steadily and evenly as possible was followed by the actual synchronization task. Participants were asked to utter the same syllables and words while synchronizing them to a series of regularly occurring tones, that were referred to as the beats of the "metronome". The IOI of beats was 750ms for the syllables, and 900ms for the wordlists. The specific instruction was to read each syllable/word to one "beep" of the metronome, starting whenever they felt ready and familiar with the rhythm. Every trial was only done once, except if there was massive misreading or severe interruptions caused by stuttering during the trial. One session lasted approximately 45 minutes. The experimenter was present during the whole session. The detailed procedure of data preparation is described in chapter 4.2. Mean accuracy (of syllables and words merged, as here no subdivision in types of stimuli is relevant) was the chosen parameter to be used for analyses as follows, since it was identified as the parameter being significantly different between PWS and PWNS in chapter 4. Accuracy represents the precision of a participant's synchronization performance: it stands for the deviation from the pacing event and can therefore be negative, in case of an anticipation or positive, in case of a delayed verbal response to the pacing event. It therefore does not tell anything about the actual variability of synchronization, but only about the distance of the response to the pacing event. For the analyses to follow, the vowel onset was chosen as the reference of synchronization measures, since it is seen as the center of the syllable/word and participants tend to synchronize their vowel to the pacing event instead of the syllable/word onset (Rathcke et al., 2021). Mean accuracy in % (of the pacing event) of all stimuli merged was chosen as a variable for the upcoming regression analyses.

Testing sessions on rhythmic parameters were scheduled following the diagnostic sessions of stuttering severity as depicted in figure 5.1. After the completion of 15 days of therapy, stuttering severity was re-evaluated with the SSI-3, (Riley, 1994) and the German Version of the OASES (Yaruss & Quesal, 2006). Again, those diagnostic sessions were done exclusively by trained and specialized speech therapists of the intensive therapy course.

	Age group	**Mean**	**Std. deviation**
SSI-3 Day 1	Young	24.36	9.34
	Old	30.35	11.99
SSI-3 Day 15	Young	16.36	8.07
	Old	11.12	9.10
OASES Day 1	Young	2.31	0.56
	Old	2.46	0.45
mean articulation rate	Young	3.50	0.79
	Old	3.67	1.07
mean accuracy	Young	2.65	3.16
	Old	3.75	3.66

Table 5.2 Descriptive statistics concerning the variables for regression analyses for all participants of both age groups

To assess therapeutic outcome, two methods being used: First, of course, the improvement in fluency was quantified by comparing pre- and post-therapy SSI-scores. Results from day 15 were chosen as criterion for the linear regression. Additionally, participants were divided into two groups depending on their post-therapy results. Since this study is not able to analyze therapeutic outcome in the long run (i.e. no follow up diagnostics of stuttering severity after some weeks or months), the division cannot be made into recovery versus persistency, as it was suggested in a prior study on this topic (Howell & Davis, 2011a). In analogy to a more recent study concerned with the factors contributing to therapeutic outcome, the term “good therapy outcome” (Cook et al., 2013, p. 126) is being used instead. SSI-3 scores of 21 or below (indicating very mild and mild stuttering) in combination with an OASES score of 2.24 or smaller (indicating mild and mild-moderate influence of stuttering) were classified as good therapy outcome. In case those conditions were not met, participants and their outcome were classified as “non-responders” to therapeutic interventions (Rozental, Andersson, & Carlbring, 2019). Despite the fact that this classification is reasonable and convenient (Cook et al., 2013) it also must be kept in mind that within this classification some participants’ changes induced by therapy might not even be ascertained in case the changes induced by therapy did not undercut the chosen values of SSI-3 and OASES – or in case their pre-therapy performance was even below those values, which does not necessarily indicate that they did not improve during therapeutic intervention – they were simply below this chosen value already displaying only mild symptoms. To account for this problem, a visual inspection of participants assignment into the groups of good therapy outcome and non-responders pre- and post-therapy is used in the following chapter (see figures 5.5a and b). All statistical analyses were conducted in SPSS (Version 27).

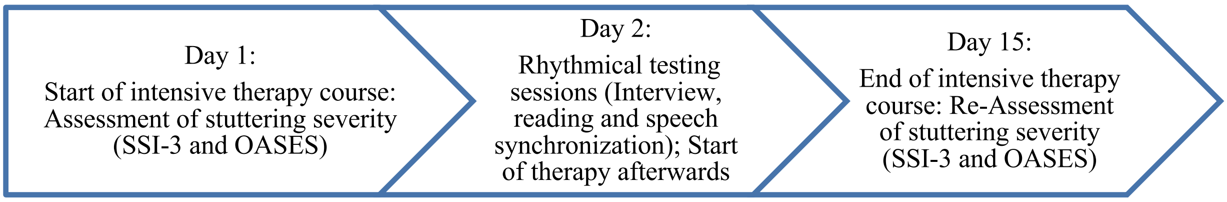


Figure 5.1 Timeline of the testing procedure

5.3 Results

Since this chapter is composed of various statistical analyses that are each quite complex, it seems reasonable to subdivide this chapter into units. Beginning with the chapter of therapy outcome analyses which quantifies therapeutic success, correlations follow to form the basis for the paragraph on regression analyses.

5.3.1 Therapy outcome analyses

After the process of data preparation and before starting regression analyses, it is important to measure therapeutic success and compare results from the SSI-3 and the OASES from the two distinct time points, since regression analyses on the factors influencing therapeutic success are only reasonable if there is a significant improvement on objective or subjective impact of stuttering. First, objective quantifications of therapeutic success are calculated: Therefore, SSI-3 values from Day 15 were subtracted from those of Day 1 to calculate a new variable containing the difference of these two values; they were controlled for normal distribution and outliers, as well. Data displayed no outliers and the differences between the pre- and post-scores of the SSI-3 were normally distributed, as assessed by the Shapiro-Wilk test (p = .051). Comparing pre- and post-values of SSI-3, results show that SSI-3 values were significantly lower after therapy with $t(38) = 7.984$, $p < .001$, $d = 1.27$ indicating a significant success of therapeutic intervention. The explorative comparison of means of pre- and post- scores of the SSI-3 emphasizes this statistically significant fact: whereas prior to therapy the mean severity was $M = 27$ (range: 7-50), indicating a moderate severity on average, the mean severity went down to $M = 14$ (range: 0-34) after therapy, indicating a mild severity of stuttering on average (see figure 5.2).

The effect of age was also taken into account when conducting the same analyses within the group of young PWS, data showed no outliers and normal distribution as well (p = .488) and a strongly significant effect of the therapeutic intervention (analyzing pre- and post-scores) with $t(21) = 4.557$, $p < .001$, $d = .92$. Within the group of older PWS, also no outliers were present, however, normal distribution of the difference of pre- and post- scores as controlled by the Shapiro-Wilk test indicated missing normal distribution (p = .017). Since paired t-tests, as chosen for these analyses are very robust, they can be applied even in cases of missing normal distribution (Stone, 2010). Furthermore, results can be compared more precisely if the chosen tests were the same. With age groups being very small – in case of older participants $n = 17$ – missing normal distribution is not surprising. Results of the paired t-test to compare pre- and post-treatment values show a significant effect of the intensive therapy course on SSI-3 scores with *t*(16) = 9.034, *p* < .001, *d* = 2.19.

It can, therefore, clearly be stated that the therapy was a success when comparing objective measures of stuttering severity as measured with the SSI-3 in a pre and post therapy.

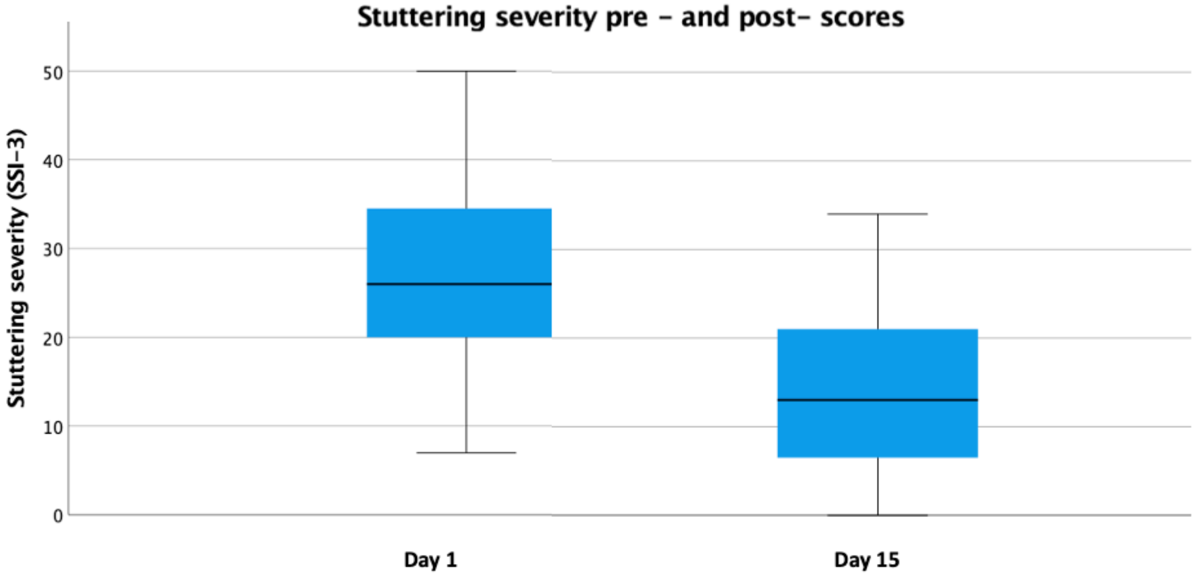


Figure 5.2 Comparison of stuttering severity pre- and post-therapy (*n* = 39)

Afterwards, subjective measures of therapeutic success were calculated: Therefore, OASES values from Day 15 were subtracted from those of Day 1 to calculate a new variable containing the difference of those values and to control for normal distribution and outliers. Data displayed no outliers and the differences between the pre- and post- scores of the OASES were normally distributed, as assessed by the Shapiro-Wilk test (*p* = .052). Results show that OASES values were significantly lower after therapy (*t*(38) = 8.045, *p* < .001, *d* = 1.28) indicating a significant success of therapeutic intervention from a subjective point of view, as well. The explorative comparison means of pre- and post- scores of the OASES emphasizes this statistically significant fact: whereas prior to therapy the mean impact of stuttering was *M* = 2.37 (range: 1.40 – 3.53), indicating a moderate impact of stuttering on the life of participants, the mean impact went down to *M* = 1.96 (range: 1.27 – 2.93) after therapy, indicating a mild-moderate impact of stuttering on average (see figure 5.3).

The effect of age was also accounted for, conducting the same analyses within the group of young PWS, data showed no outliers and normal distribution as well (*p* = .675) and a strongly significant effect of the therapeutic intervention when comparing pre- and post-therapy values (*t*(21) = 6.60, *p* < .001, *d* = 1.42). Within the group of older PWS, also no outliers were present and normal distribution of the difference of pre- and post- scores as controlled by the Shapiro-Wilk test indicated that normal distribution was given (*p* = .087). Results of the paired t-test (pre- and post-therapy values) show a significant effect of the intensive therapy course on OASES scores with $t(16) = 4.644$, $p < .001$, $d = 1.126$.

It can, therefore, clearly be stated that the therapy was a success, not only when comparing objective measures of stuttering severity as measured with the SSI-3, but also when comparing subjective impact rates in a pre- and post- design.

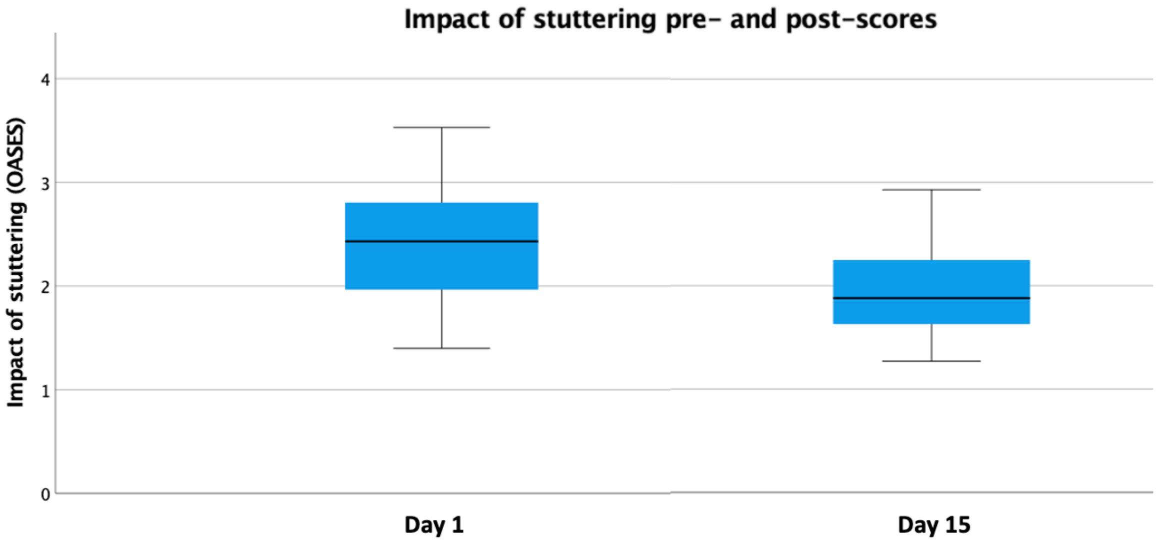


Figure 5.3 Comparison of the impact of stuttering on the life of participants pre- and post-therapy (*n* = 39)

Furthermore, the division of participants into the two groups of good therapy outcome and non-responders – as described in chapter 5.2.2 is visualized below in the pie charts in figure 5.4. Here, of course, no comparison of pre- and post- values is possible since values are only based on therapy outcome values of SSI-3 and OASES. However, it is interesting to look at the figures for young and old participants, since here it becomes evident that the effect is carried by young participants exclusively, as more than half of the older participants are classified as non-responders. However, group comparisons of SSI-3 and OASES results pre- and post- therapy reached significance for both groups merged and age groups separately, indicating that this effect can be seen as a trend but not a statistically significant difference concerning age groups, which furthermore only applies to the combination of SSI-3 and OASES post therapy within the term of therapy outcome – as results on SSI-3 and OASES values post-therapy have proven to be significantly lower in both age groups. As younger participants have had lower initial stuttering severity in general, this might explain the observed effect in figure 5.4.

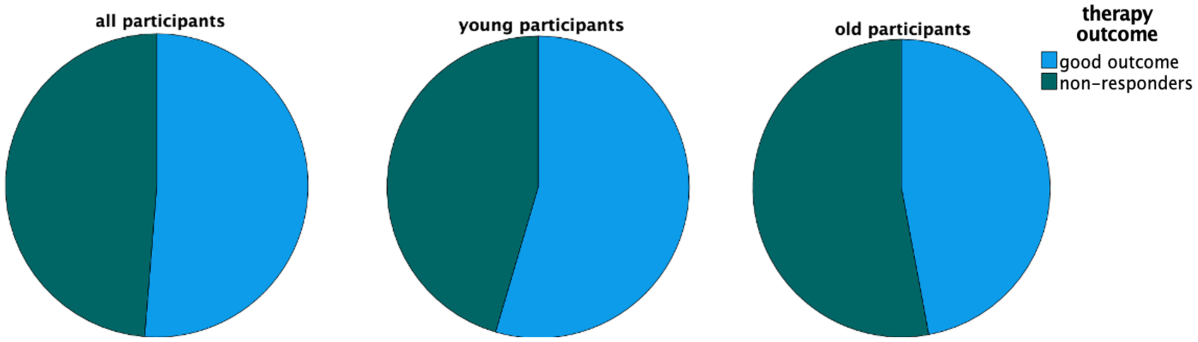


Figure 5.4 Visualization of therapy outcome in pie charts

As the definition of good therapy outcome has clear cut-off values for good outcome versus non-respondent, individual improvements might not be taken into consideration. Since participants with higher initial severity who also show big improvements might still not be below the cut-off values defined, whereas participants with lower initial severity do not have to improve that much to fall below the defined cut-off value, this way of defining therapeutic outcome also displays disadvantages. Still, this option seemed more reasonable than using the mere improvement as calculated by subtracting values from day 15 from those of Day 1, as those raw numbers are based on an ordinal scale which is not suited for direct comparisons of improvement. Furthermore, there is to date no literature on values defining good therapy outcome versus non-respondent to therapy (Cook et al., 2013). The chosen way of defining therapy outcome, therefore, seems like the more suitable option and is supported by the visualization of improvement in SSI-3 and OASES values showing that there is a clearly visible improvement with a change in categories (as indicated by the red line in graphs 5.5a and b) when inspecting the mean values of SSI-3 and OASES pre- and post- therapy. As can be seen in figure 5.5b, some of the initial OASES values were low enough to fall under the cut-off value, however, for SSI-3 values there is no overlap and hence a clear distinction between categories.

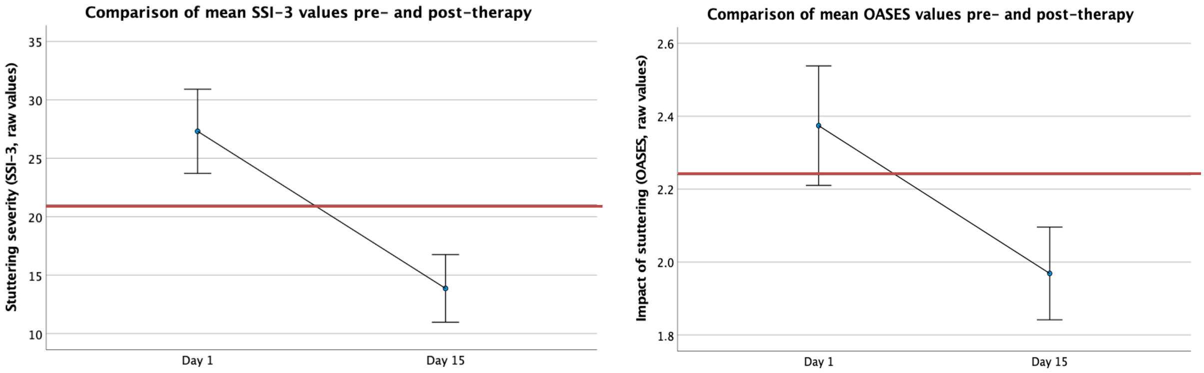


Figure 5.5a and b Mean values of objective and subjective stuttering severity from pre- and post-therapy with the red line indicating the cut-off value for the classification of therapy outcome.
Error bars represent 95% confidence interval.

It has been demonstrated powerfully that 15 days of intensive therapy had strong impact on subjective impact of stuttering on the participants life as well as on objective measures of stuttering severity. This raises the question about the potential parameters leading to this success. The following regression analyses will clarify this question by determining the contribution to this success of the following parameters: initial stuttering severity as measured by SSI-3 and subjective impact of stuttering on the participant's life as measured by the OASES, mean articulation rate (of speaking and reading merged) as well as parameters of verbal sensorimotor synchronization, namely the mean accuracy of synchronization. As within chapter 4 many interesting parameters of verbal synchronization were investigated (i.e. variability, consistency) mean accuracy was chosen for the upcoming regression analyses for two reasons: on the one hand, predictors must be limited to a certain number depending on the total number of participants, on the other hand, accuracy was the parameter displaying the biggest differences between groups and therefore was considered to be most promising in having a predictive power.

5.3.2 Correlations

Prior to regression analyses, correlations between the data collected for the upcoming regression analyses are conducted to determine possible significant relationships and to identify potential predictors. Pearson's correlation coefficient *r* is used comparing the following data: SSI-3 pre- and post- therapy, therapy outcome (i.e. good therapy outcome versus non-responders), OASES Day 1, mean articulation rate and mean accuracy in verbal synchronization. As regression analyses will not being conducted for age groups separately due to reasons of sample size, correlations will not be conducted for both groups either.

Furthermore, chapter 5.3.1. has demonstrated that effects of therapy reached significance – independently of the group constellation (groups merged versus separated). All correlations are summarized in table 5.3. However, scatter plots are used to see if there are any big differences concerning age groups in the following correlations. As therapy outcome is a dichotomous variable, scatter plots are not useful here and therefore not shown. However, visualizations of other significant correlations might help to better understand these relations.

SSI-3 pre therapy showed a significant relationship with SSI-3 post therapy ($r = .507$, $p = .001$) and with mean articulation rate ($r = -.567$, $p < .001$). The correlation of SSI-3 pre- and SSI-3 post-therapy indicates, of course, that lower outcome values are linked to lower initial stuttering severity. This effect seems to be stronger in old participants, as figure 5.6a visualizes. Furthermore, the line with gradient = 1 demonstrates powerfully that besides four data points all other data points were above the line, indicating a good respondence to therapy. This visualization in plots underlines the preceding figures on therapy outcome as it clearly shows the high number of data points above the line, which stand for a therapeutic success in form of lower SSI-3 values. The negative correlation of SSI-3 pre therapy with mean articulation rate indicates that initial higher severity of stuttering is accompanied by lower a lower mean articulation rate. Here a tendency becomes visible, hinting towards the fact that old participants display a higher severity and lower rates 5.6b.

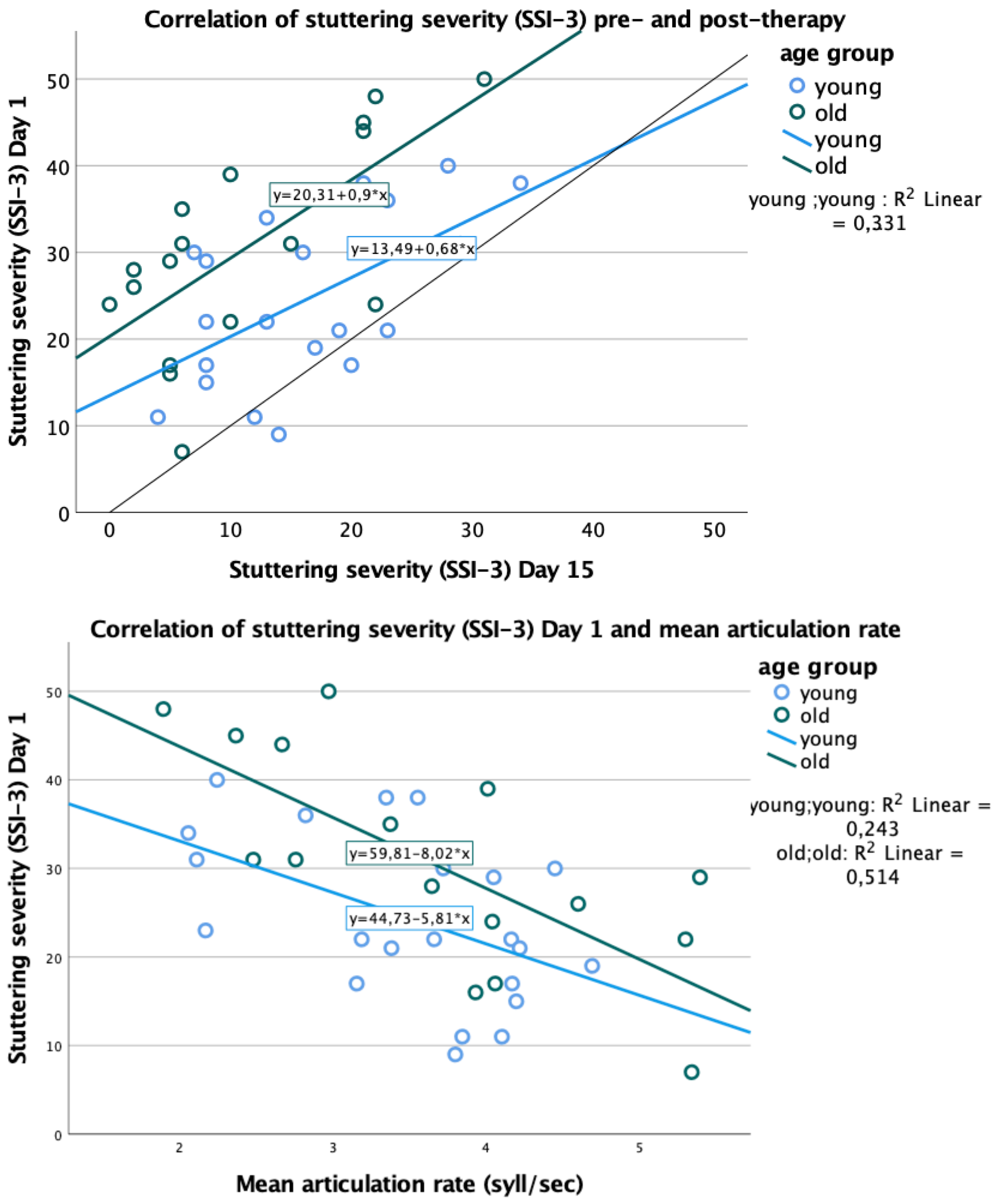


Figure 5.6a and b Correlations of SSI-3 pre- and post-therapy and SSI-3 pre-therapy and mean articulation rate:

Figure 5.6a includes a line with a gradient of 1 to help visualize change from pre- to post-therapy. The more points lie above the line the greater the improvement from Day 1 to Day 15 (points on the line indicate no change).

Furthermore, SSI-3 post-therapy values show significant correlations with mean articulation rate ($r = -.556$, $p < .001$, see figure 5.7). Correlations of SSI-3 post therapy and articulation rate indicate that lower rates were associated with more severe symptoms after therapy. Here no clear trend of age groups is visible.

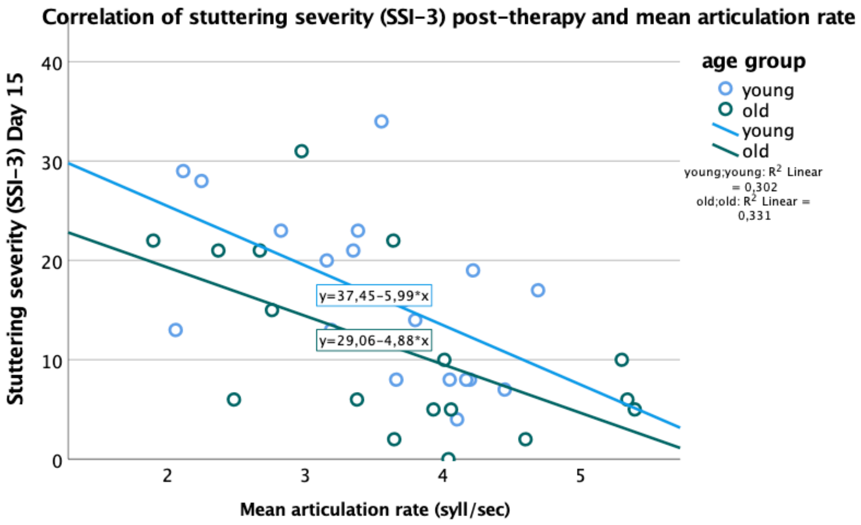
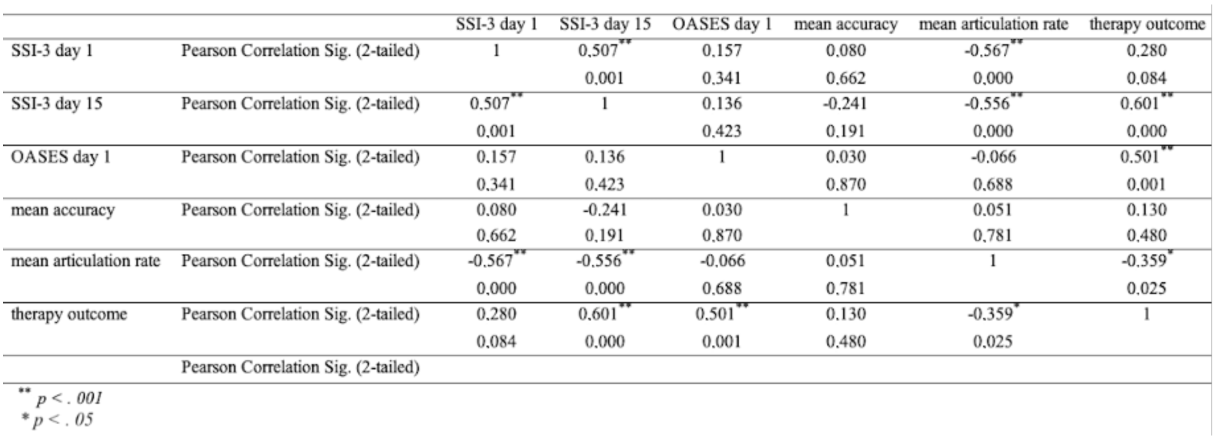


Figure 5.7 Correlation of SSI-3 post-therapy and mean articulation rate

OASES values correlated significantly with therapy outcome (*r* = .501, *p* = .001), as the values of OASES were part of the calculation of therapy outcome and therefore show a relationship. Therapy outcome correlated significantly with SSI-3 after therapy (*r* = .601, *p* < .001), OASES (*r* = .501, *p* = .001), and mean articulation rate (*r* = -.359, *p* = .025). The first correlation can be explained by the calculation of therapy outcome, that contains SSI-3 post therapy values as well as OASES values within this calculation. The correlation of mean articulation rate and therapy outcome indicate that a lower rate is associated with being classified as non-responder as outcome.



		SSI-3 day 1	SSI-3 day 15	OASES day 1	mean accuracy	mean articulation rate	therapy outcome
SSI-3 day 1	Pearson Correlation Sig. (2-tailed)	1	0.507**	0.157	0.080	-0.567**	0.280
			0.001	0.341	0.662	0.000	0.084
SSI-3 day 15	Pearson Correlation Sig. (2-tailed)	0.507**	1	0.136	-0.241	-0.556**	0.601**
		0.001		0.423	0.191	0.000	0.000
OASES day 1	Pearson Correlation Sig. (2-tailed)	0.157	0.136	1	0.030	-0.066	0.501**
		0.341	0.423		0.870	0.688	0.001
mean accuracy	Pearson Correlation Sig. (2-tailed)	0.080	-0.241	0.030	1	0.051	0.130
		0.662	0.191	0.870		0.781	0.480
mean articulation rate	Pearson Correlation Sig. (2-tailed)	-0.567**	-0.556**	-0.066	0.051	1	-0.359*
		0.000	0.000	0.688	0.781		0.025
therapy outcome	Pearson Correlation Sig. (2-tailed)	0.280	0.601**	0.501**	0.130	-0.359*	1
		0.084	0.000	0.001	0.480	0.025	
	Pearson Correlation Sig. (2-tailed)						

** *p* < . 001
* *p* < . 05

Table 5.3 Correlations conducted for all participants (*n* = 39) with the first line showing the correlation coefficient *r* (marked if significance was reached) and the second displaying the exact *p*-value

5.3.3 Regression analyses

The correlations calculated in chapter 5.3.2 revealed that predictors correlate with each other as well as with outcomes. For the detailed assessment of the relation between the named variables a linear regression analysis is carried out. SSI-3 values from Day 15 (after therapy) are used as the outcome variable.

Since therapy outcome (good outcome versus non-responding) is a binary variable, a logistic regression was carried out afterwards to investigate the improvement in fluency (SSI-3 post-therapy) and the subjective perception of participant's stuttering (OASES post-therapy).

5.3.3.1 Linear regression analysis to predict SSI-3 after therapy

For the first regression analysis investigating the outcome of therapy as measured by SSI-3 (Day 15), the following predictors were used: SSI-3 Day 1, OASES Day 1, mean articulation rate and mean accuracy.

The model has no autocorrelation as the value of the Durbin-Watson statistic is 1.844. Data were checked for multicollinearity, indicating there is no multicollinearity between predictors as controlled for with tolerance/VIF (all values between 1.0 and 1.7). Linearity and homoscedasticity were controlled for by visually inspecting the scatter plot of standardized residuals against standardized predicted values which displayed a random and even dispersion throughout the scatter plot and therefore indicated that the criteria of linearity and homoscedasticity were met. To check for normal distribution of the data, the histogram and the normal probability plot for the standardized residuals were inspected as well, indicating a normal distribution. The R^2 for the overall model was .483 (adjusted R^2 = .404), indicative for a high goodness-of fit according to Cohen (1988). SSI-3 Day1, OASES Day 1, mean articulation rate and mean accuracy were able to predict SSI-3 Day 15 statistically significantly (F (4, 26) = 6.075, p = .001). With the model at hand, 40.4 % of stuttering severity after therapy as measured with SSI-3 can be explained with the only significant predictor, however, being mean articulation rate with a regression coefficient of – 4.471 and p = .019. As table 5.4 shows, all other variables (mean accuracy: p = .098; SSI-3 day 1: p = .201; OASES day 1: p = .403) lacked significance and are therefore not suited to predict SSI-3 after therapy.

With the coefficient of determination R^2 = .483, n = 39 and α = .05, the model with four predictors has a statistical power of 1- β = .998 (Hemmerich, 2019). The model can be considered a good representation of the data, since only one participant (namely P15) had a standardized residual bigger than 2 (precisely 2.494) and therefore stayed below the critical mark of 5 % of the data with the one case representing 2.56 % in total (Field, 2009).

	non standardized coefficients		standardized coefficients		
	regression coefficient	*SE*	beta	T	*p*
Constant	21.765	11.087		1.963	0.060
mean accuracy	-0.627	0.365	-0.245	-1.715	0.098
mean articulation rate	-4.471	1.780	-0.452	-2.512	0.019
SSI-3 day 1	0.196	0.149	0.237	1.312	0.201
OASES day 1	1.972	2.320	0.121	0.850	0.403

Table 5.4 Influence of all coefficients on the dependent variable SSI-3 day 15 with significance values

After the detailed description of the chosen model, an interesting correlation confirms what has been shown already: the prediction of the model can be considered a solid and precise calculation, as the correlation (Pearson's correlation coefficient *r)* of predicted and actual stuttering severity as measured with SSI-3 is strong ($r = .685$, $p < .001$) and the visualization in the scatter plot (figure 5.8) shows a linear relationship. A higher predicted SSI-3 post-therapy value hence was associated with a high SSI-3 value as diagnosed after therapy. This finding further supports the result of the linear regression analyses which was able to predict SSI-3 values post-therapy statistically significantly (F (4, 26) = 6.075, $p = .001$).

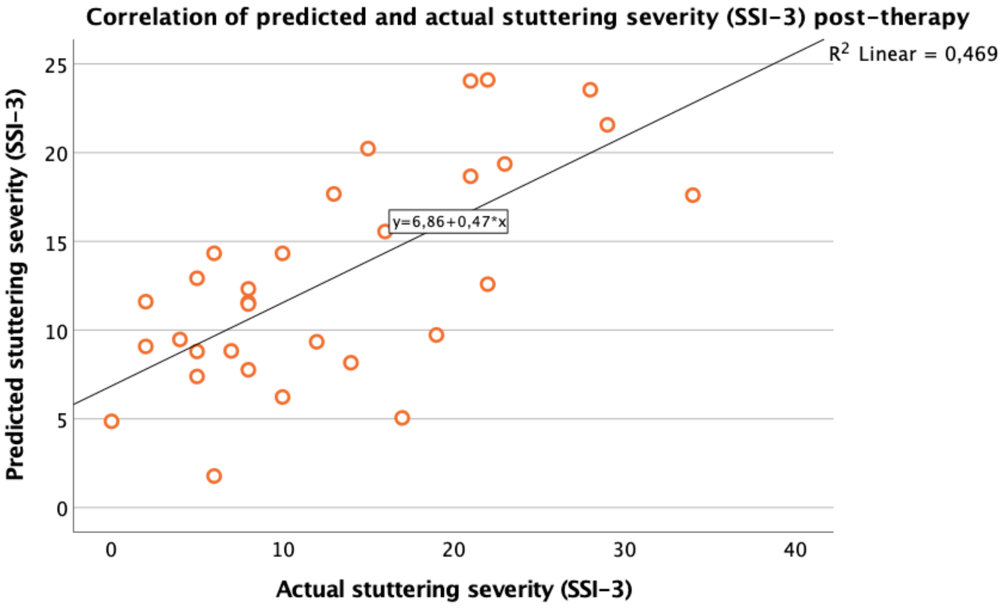


Figure 5.8 Correlation of predicted and actual stuttering severity (diagnosed with SSI-3) post-therapy

After results of the linear regression, these analyzes will be complemented by a logistic regression to investigate the probability of a good therapy outcome.

5.3.3.2 Logistic regression to predict therapy outcome

Logistic regression analysis was used to determine if it is possible to predict the probability of a good therapy outcome versus non-responders in patients based on the information from the initial assessment. This information is highly relevant when it comes to the prognosis made when setting goals for an intensive therapy ahead.

Logistic regression was therefore employed using the same coefficients, namely mean accuracy, mean articulation rate, SSI-3 Day 1 and OASES Day 1 with the differences concerning the outcome measure: not only a number to quantify objective stuttering severity but a combined measure of objective und subjective therapeutic outcome that has the two dichotomous outcome possibilities of good therapy outcome and non-responders. This measure might be of even higher relevance for patients themselves since it is not always the mere severity of stuttering that has the most influence on daily life and the perception of the impact of stuttering on one's life, but rather the combination of fluency during speaking and the feelings of a participant towards his own stuttering and therapeutic success. Therefore, a logistic regression was chosen to answer the question if measures collected prior to therapy have the power to predict therapy outcome that is defined in a more comprehensive way than the mere severity.

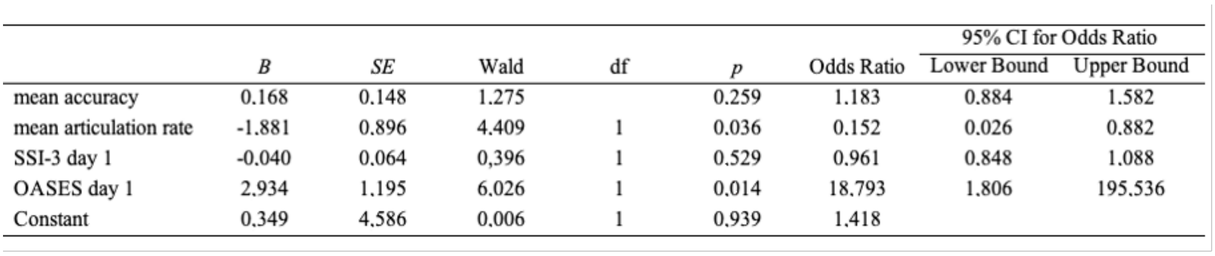
In a first step, all independent variables included were logit transformed according to the Box-Tidewell Transformation (Box & Tidewell, 1962) in order to test linearity. Bonferroni-correction was then applied to all eight terms in the model (Tabachnick & Fidell, 2018). All variables displayed a linear relationship. Correlations between predictor variables indicated that multicollinearity is not a confounding factor in the conducted analysis since all correlations were low ($r < .70$). Data were controlled for outliers using the studentized residuals: according to the definition of the cut-off value of 3 (Pardoe, 2012, p. 166; Yan & Su, 2009, p. 134), two participants were classified as outliers, namely P114 (SResid = 2.124) and P123 (SResid = -2.781). According to Field (2009), a model is still considered to be a good representation of data if less than 5% of the data display residuals with values greater than 2. In case of the thesis at hand, the two cases represent 5.12% and are at the border of this cut-off value. Therefore, a second way to identify outliers, the Cooks Distance was added: Here two cases were found, as well. However, it was different participants (P80 and P71) who exceeded the value of 1 which is recommended as cut-off value for the classification of outliers (Heiberger & Holland, 2015,

p. 367). As the classification of outliers led to different results and they are only few in number, the model will continued with the outliers included, knowing that in case of inconclusive results, the model can be re-calculated with outliers excluded.

A binomial logistic regression was performed to determine the effect of mean accuracy, mean articulation rate, initial stuttering severity and subjective influence of stuttering on a participant's life and predict the likelihood of good therapy outcome versus non-responding. The chosen binomial logistic regression model was statistically clearly significant, χ^2 (8) = 22.086, p = .005, as a large amount of variance (Backhaus, Erichson, Plinke, & Weiber, 2006, p. 456) is being explained by this model which is shown by Nagelkerke's $R^2 = .557$. Goodness-of-fit was assessed as well, using the Hosmer-Lemeshow-Test, which indicated a good model fit χ^2 (8) = 6.005, p > .05 (= .647) and Cox and Snell with $R^2 = .417$. With a sensitivity of 88.2% and a specificity of 86.7% the overall percentage of accuracy in classification was 87.5% and can therefore be considered a high percentage of accuracy in classification.

Of the four variables entered into this regression model, two displayed a significant contribution in predicting therapeutic outcome when employing a .05 criterion of statistical significance, as it was done in all analyses before: mean articulation rate ($p = .036$) and OASES Day 1 ($p = .014$), while the other two variables showed no significant effect: mean accuracy ($p = .259$) and SSI-3 day 1 ($p = .529$). Mean articulation rate had an effect on showing a good therapy outcome versus being non-responder, OR = .152 (95% CI[.026, .882]), as did OASES Day 1 OR = 18.793 (95% CI[1.806, 195.536]). Interpreting these results in detail, mean articulation rate displays a negative regression coefficient (-1.881), indicating that a rise in articulation rate of a single unit has a negative influence on the probability of being classified as non-responder. Using the odds ratio of .152 and calculating the percentage it becomes evident that a rise of articulation rate of a single unit makes the odds of being in the group of non-responders decrease by 84.8%. To put it in simpler words: a higher articulation rate is rather associated with a good therapy outcome instead of being non-responder. For OASES values of day one, the second significant predictor, regression coefficient was positive (2.934), indicating that a rise in OASES values of a single unit has a positive influence on the probability of being classified as non-responder. Using the odds ratio of 18.793 and calculating the percentage it becomes evident that a rise of OASES values of a single unit makes the odds of being in the group of non-responders rise by 17.79%. This means that higher values of OASES Day 1 are associated with no respondence to therapy as outcome.

Table 5.5 shows all model coefficients and odds as well as the beta values (regression coefficient) and their standard errors, the Wald statistic and significance values.



	B	*SE*	Wald	df	*p*	Odds Ratio	95% CI for Odds Ratio	
							Lower Bound	Upper Bound
mean accuracy	0.168	0.148	1.275		0.259	1.183	0.884	1.582
mean articulation rate	-1.881	0.896	4.409	1	0.036	0.152	0.026	0.882
SSI-3 day 1	-0.040	0.064	0,396	1	0.529	0.961	0.848	1.088
OASES day 1	2.934	1.195	6.026	1	0.014	18.793	1.806	195.536
Constant	0.349	4.586	0.006	1	0.939	1.418		

Table 5.5 Logistic regression predicting therapeutic outcome

A comparison of the initial model with only the constant in the regression equation predicting 53.6% of participants correctly (and therefore being only slightly above chance) and the final model which predicted 87.5% of participants correctly after adding the four predictors shows that the knowledge about the chosen predictors improved results drastically (Reed & Wu, 2013).

5.4 Discussion

The goal of this study on regression models was to find out about the potential that lies in rhythmical abilities and temporal parameters as predictors for therapeutic outcome and to gain more information on factors influencing therapeutic outcome in terms of objective stuttering severity and good outcome versus non-respondence to therapeutic intervention. To do so, thirty-nine German-speaking children and adolescents were tested before and after an intensive therapy course lasting 15 days. Data collection prior to therapy included stuttering-specific diagnostics (SSI-3 and OASES) as well as rhythmical test (articulation rate and accuracy of verbal synchronization). After the intervention, data on stuttering severity (SSI-3 and OASES) were obtained again. To evaluate the predictive power of the chosen values, two different outcome measures were used: firstly, outcome in fluency as measured by SSI-3 after therapy, which was evaluated by linear regression and secondly therapy outcome, as measured by a combination of SSI-3 and OASES values post-therapy, which was evaluated by using logistic regression analyses.

Results will be discussed regarding the hypotheses postulated at the beginning of this chapter: Linear and logistic regression analyses partly confirm hypotheses and partly lead to the rejection of hypotheses. Firstly, it was assumed that participants with a high initial stuttering severity as measured with SSI-3 on Day 1 and OASES Day 1 rather won’t have a good therapy outcome. However, neither the linear, nor the logistic regression analyses could confirm this hypothesis, as the initial SSI-3 value did not reach significance as predictor for SSI-3 on Day 15 or for therapy outcome. However, OASES values of Day 1 reached significance in the logistic regression analysis, being a significant predictor for therapy outcome. This hypothesis, therefore, must be rejected in terms of SSI-3, but can be confirmed for OASES values within the logistic regression analysis. Secondly, it was hypothesized that participants with a mean articulation rate which is closer to rates of fluently speaking participants (i.e. higher) were more likely to show a better outcome in terms of SSI-3 values post therapy and therapy outcome. This hypothesis was confirmed for both analyses, the linear and the logistic regression, since mean articulation rate turned out to be the only predictor that was significant for both analyses. Finally, mean accuracy on the verbal synchronization task was expected to influence therapy outcome and therefore the hypothesis was postulated that a more accurate performance would also lead to a better outcome after therapy in terms of lower SSI-3 values and a good therapy outcome. However, this hypothesis is to be rejected completely, as neither for the linear regression analysis, nor for the logistic regression analysis was this predictor able to reach significance. In sum it can be stated that some of the hypotheses could be confirmed as they showed the expected results, others, however, must be rejected as they could not be confirmed in this study.

Discussing the results regarding the current literature on this topic, it was quite surprising for the data at hand, that SSI-3 values pre therapy did not turn out to be significant predictors, as many other studies identified initial stuttering severity (also diagnosed with SSI-3) as a significant predictor for the outcome of therapy and found out that participants with an initial higher severity "[…] were more likely to persist […]" (Cook et al., 2013, p. 131). Further authors (Block, Onslow, Packman, & Dacakis, 2006; Howell & Davis, 2011a) also identified higher SSI-3 values as a significant predictor of persistent stuttering. It must be stated, however, that when quantifying therapeutic outcome, the moment of investigation can vary significantly influencing results strongly, as some studies chose to re-evaluate stuttering severity right after the therapeutic intervention (see Cook et al., 2013), whereas others defined therapeutic success in the long run with re-evaluations taking place several years after treatment (Block et al., 2006). This variation can of course lead to results that can barely be compared directly.

Looking at the impact of stuttering on participants' life (as measured with OASES), a comparable study by Cook and colleagues (2013) did not identify a questionnaire on the psychosocial impact of stuttering, namely the FzS (Cook, 2013) as a significant predictor. However, a significant correlation of FzS values and the improvement in fluency was found, indicating a connection between these two measures (Cook et al., 2013). The fact that OASES

Day 1 values were found to be a significant predictor for the logistic regression analysis in the thesis at hand is not surprising considering those facts: OASES represents the impact of stuttering on participants' everyday life and can, therefore, be seen as measurement of subjective stuttering severity as well, since it reflects every person's very own feelings about his or her stuttering and how it impacts daily situations (Yaruss & Quesal, 2008). For an encompassing diagnostic on stuttering severity this aspect of the disorder also must be taken into account and as objective stuttering severity proved to be an important predictor in the studies cited above, subjective stuttering severity consequently also can have the power to predict therapy outcome. It was already in 1976 that Guitar found pre-treatment attitudes towards stuttering to be highly related to therapy outcome (Guitar, 1976). It therefore is not surprising to find OASES values also representing a person's attitude to be a significant predictor for therapy outcome. Furthermore, the logistic regression analysis of the present paper used the term of good therapy outcome, which was composed of SSI-3 and OASES values post therapy. It therefore is quite logical that initial OASES values show impact on OASES values conducted post therapy as there is a direct connection between these evaluations.

Articulation rate, as a very basic form of speech rhythm, displayed strong predictive power as it was the only variable that reached significance in both analyses. Already in 1999 authors specialized in stuttering (Hall et al.) investigated the potential prognostic power of articulation rate: comparing articulation rate in perceptually fluent utterances of CWS, Hall et al. (1999) found out a clear tendency for children who later recovered from stuttering to display slower rates than persisting children and, therefore, suggested to use it as an indicator for persistent versus recovered stuttering. Although results failed to reach significance a tendency was visible when comparing data on articulation rates. However, the direction found in this study by Hall (1999) differs from the results at hand: whereas Hall and colleagues (1999) found rates in later recovered PWS to be slower compared to persisting participants, the thesis at hand found out that a higher rate (and therefore closer to the rate of PWNS) was associated with good therapy outcome. It must be noted, though, that the outcome definition was not the same, as the thesis at hand did not use recovered stuttering but only good therapy outcome. A further investigation by Kloth et al. (1999) examined fluent utterances of children at two distinct points: before stuttering onset and then again, one year after onset. They found a higher variability concerning the articulation rate in persisting children than in recovered children – at both moments of investigation (Kloth et al., 1999). A higher variability cannot be compared directly to the results at hand, they do, however, also point towards the fact that recovered children tended to show an articulation rate that resembled more the one of fluently speaking children, which is in line with the results above. A further study interested in the link between stuttering severity and articulation rate came to the results that a slower articulation rate in CWS was associated with a higher frequency in stuttering-like disfluencies, and longer durations of prolongations (Tumanova et al., 2011). This would be in line with the results of the study at hand in terms of PWS displaying slower rates and the fact that good therapy outcome was associated with higher rates. However, this study did not investigate any predictive character of articulation rate and was conducted with pre-school-aged children. Therefore, results are not perfectly comparable. Investigating articulation rate in a comparable age-span, Erdemir et al. (2018) conducted a study with children from three to five years. They were subdivided into groups of persisting, recovered and nonstuttering children. Faster articulation rates within fluent speech were found in recovered compared to persisting children. These results go along with the thesis at hand.

Regarding accuracy of synchronization, which represents a very typical form of rhythmic investigation within the population of PWS, to date no study is known that investigated the prognostic power of rhythmic synchronization to differentiate PWS in groups of persisting versus recovered stuttering or non-responders versus good therapy outcome. Furthermore, no research on the influence of rhythmic abilities on stuttering severity after an intensive therapeutic intervention had been conducted, so far. Despite many results supporting the hypothesis of atypical rhythm in stuttering (Ladány et al., 2020) and the fact that rhythm perception or production were already used to predict other abilities, such as reading outcome in developmental dyslexia (Flaugnacco et al., 2014) or related to other cognitive, linguistic or perceptual skills (Tierney & Kraus, 2013), the specific usage of verbal rhythm in stuttering outcome research has not been investigated so far. Merely the fact that rhythmic skills in PWS have not been used as therapy outcome predictors yet, but also the vast amount of literature on the topic of atypical rhythm in PWS of any age and in many domains (Chang et al., 2016; Falk et al., 2015; Olander et al., 2010; Wieland et al., 2015) turn this investigation into a highly interesting one. Despite accuracy in verbal synchronization differing significantly between PWS and PWNS as demonstrated in chapter 4, mean accuracy did not reach significance as a predictor in any of the two regression analyses. This result seems surprising at first, as accuracy in verbal synchronization did not only display significant differences in chapter 4 but is a rhythmic construct, just as articulation rate. It also must be considered that it is a very complex task indeed. It is possible that the complexity of this task may have layered the fact that this rhythmic task might have the power to predict therapy outcome; perhaps another task within the spectrum of rhythmic synchronization (non-verbal, i.e. tapping or clapping; perceptive instead of productive, i.e. rhythm discrimination; different stimuli, i.e. musical rhythm instead of metronome pacing) would have had the power to do so. This question, however, cannot be solved within the present paper but it is a topic for future research in this very promising field. Furthermore, the rather small sample of n = 39 makes it harder to detect systematic differences between populations. Hence, a bigger number of participants with a more homogenous age profile would be desirable for further investigations, as well.

Some further limitations of the thesis at hand must be discussed: firstly, a logistic regression was used to investigate the influence of predictors on a dichotomous outcome event – in this case good therapy outcome versus non-responders. However, as this outcome event is treated as dichotomous, it might as well be argued that it is a rather continuous outcome that has been made into a dichotomous one by defining a cut-off criterion. It can be argued that this splitting of outcome into good therapy outcome versus non-responders is not dichotomous per se, unlike dichotomous events that are unarguably categorized into classes of having occurred and not having occurred (e.g. the occurrence of a bone fracture). The dichotomous outcome of the logistic regression in this study is rather comparable to the presence or absence of pathologically high blood pressure, where values are continuous and a cut-off defines which values are above the normal range (Reed & Wu, 2013). However, this approach is in line with stuttering specific literature on therapy outcome that was published in an international, peer-reviewed journal (Cook et al., 2013) and cut-off criteria were chosen wisely and reasonably. A further limitation is – of course – sample size. With $N = 39$ sample size is considered small and clearly below the recommended minimum numbers of around 250 participants for logistic regression (Reed & Wu, 2013). Results of studies with small numbers are not to be ignored, but there is a risk that results might have over-estimated the amount of influence of the factors chosen for the outcome prediction. The calculation of logistic regression can furthermore be affected by small sample sizes in terms of overestimated odds ratios caused by inherent properties of these kind of regression models. This was also a reason why regression analyses were not conducted for age groups separately, since – besides the interest in the information this analysis might provide – it could not be interpreted without great caution due to the very small numbers included when separating the complete sample into the two samples for age groups.

Another problem that must be addressed is the class imbalance problem, meaning that when trying to predict therapeutic outcome in terms of good therapy outcome or even recovery versus persistency, this outcome does not only depend on the quality of therapeutic intervention or rhythmic skills of participants, but also simply on the fact that within younger participants the rate of recovery is higher than within older participants. In case of the study at hand, a subdivision of participants in younger versus older for the calculation of regression models would not have been effective, since sample size would have reduced drastically. Instead, a case balanced model for predicting therapy outcome was computed to account for the problem of sampling imbalances.

When it comes to selecting potential predictors, it surely is not the best idea to simply use as many as possible – but rather the opposite seems to be true. Choosing predictors selectively and wisely towards the research question at hand is important, with the rule of 10 participants per variable studied – at least for logistic regression models (Agresti, 2007; Reed & Wu, 2013). This obviously leads to constraints within the chosen method; however, the thesis at hand tried to stick to this rule by choosing 4 predictors.

Therapeutic effects after an intensive therapy course such as the one in the three studies conducted are quantified right after the end of the treatment. At this point, success in terms of fluency is highest and impact of stuttering is mostly lowest, since the motivation to use all the freshly learned techniques is high and the knowledge how to use them properly is still very present (Guitar, 2014; Thum & Mayer, 2014). So on the one hand it seems very reasonable to use diagnostic values from the day right after treatment ended as criterion; on the other hand it must also be considered that changes on neurological levels as they are observed after successful therapy (i.e. neuroplasticity) will most likely take more than just the 15 days of intervention. After a complete recovery from stuttering, an activation within the left orbitofrontal cortex and in the auditory cortex bilaterally was found. Those results after effective therapy or recovery all hint towards a normalization of neurofunctional activation, just as it is common in fluently speaking persons (Lu et al., 2012; Lu et al., 2017). However, such complex processes will not have taken place during 15 days of therapy, even if the outcome is really good in terms of fluency and attitudes towards stuttering. Furthermore, the timespan of 15 days of therapy, even if it is an intensive therapy course, is still quite small and results might have turned out even better (i.e. lower SSI-3 values, lower OASES values) if the intervention had lasted longer, comparable to the study conducted by Cook et al. (2013).

A further consideration is the therapy type of this study: as method combination does not primarily aim at a high level of fluency whilst speaking, but rather at the possibility of the person speaking to choose the manner of speaking, results might have turned out differently when using other types of therapy. A fluency shaping therapy with the goal of high speech fluency might have been influenced differently by the predictors taken into account in this chapter. At the same time, a stuttering modification therapy, which does not aim at high levels of fluency but rather at the feeling of being in control of one's own speech also would have shown different results, as probably the therapy outcome itself would have been different with

SSI levels not as low as in modification. As to date there is no study comparing predictors for each type of therapy, these ideas remain speculative.

Despite all the limitations just discussed these results also provide promising results for further investigations in the field of outcome predictors. Even though the classical rhythmic task (i.e. verbal synchronization) did not turn out as a significant predictor, a rather basic form of rhythm, articulation rate, did. Considering the effort of obtaining the value of mean accuracy compared to a mean articulation rate, this proportion even suggests using articulation rate instead of values obtained in verbal synchronization tasks: whereas articulation rate can be ascertained by using the spontaneous speech and reading sample which is needed for the SSI-3 pre therapy anyways, tasks of verbal synchronization are quite complex to develop and conduct, with lots of technical devices needed. Also processing and analyzing verbal synchronization data is a lot more time consuming than the evaluation of articulation rate (see chapter 3 and 4 for details in the process). Therefore, articulation rate seems like the more feasible option, as the basis for obtaining it is also part of the mandatory implementation of SSI-3 in a diagnostic session and analysis is much faster and easier. OASES (or other questionnaires used to determine influence of stuttering on a person's life) pre-treatment values are obtained in nearly all therapies anyways and they are very easy to analyze, therefore, it can be considered a great benefit that OASES values could also be used to predict therapeutic outcome.

Whilst verbal synchronization data did not turn out as a significant predictor for therapeutic outcome, it might still be worth testing those rhythmic parameters in bigger populations or slightly different tasks. Besides its potential power to be used as a tool in outcome prediction, rhythm and temporal parameters also entail huge potential as a therapeutic element. Since rhythm in general or singing in particular has proven to be a great therapeutic tool in other disorders like Parkinsons's disease (Benoit et al., 2014; Cochen De Cock et al., 2018), this approach is worth trying in a disorder which displays so many links to rhythm – not only in terms of atypical rhythm but also in terms of its positive and fluency enhancing effects. Future research still has a wide field to discover and many facets of rhythm in stuttering to be explored.

6. General discussion

This thesis aimed at investigating rhythmic skills and temporal parameters in a population of children and adolescents who stutter and comparing them to skills of typically fluent peers in order to carve out specific characteristics of rhythmic profiles in PWS. The investigation of these rhythmic skills was limited specifically to the domain of speech rhythm found in fluent parts of speech. The main goal was to gain more insight into ongoing processes of speech production and speech motor execution in order to contribute to possible explanations for the occurrence of stuttering symptoms within the aberrant systems of speech processing and production within PWS. Furthermore, the newly gained information on speech rhythmical skills was investigated in terms of how suitable it is for the prediction of the therapeutic outcome. Chapter 3 examined a very basic form of speech rhythm or rather temporal parameters, namely the speech and articulation rate, with the finding that both speech and articulation rate differed significantly between PWNS and PWS. Chapter 4 investigated a more specific form of speech rhythm, the so-called sensorimotor synchronization, with a verbal synchronization paradigm. The results indicated that rhythmically cued and perceptually fluent speech of PWS is still very different from the speech of PWNS in terms of accuracy. In chapter 5 it was examined whether those exact parameters that were found to deviate so clearly in PWS also had the power to predict the therapeutic outcome of an intensive therapy. In this final chapter, all results are summarized in detail, related to other recent findings in this field and the relevance of these results in terms of stuttering diagnostic and therapy will be demonstrated.

6.1 Summary of the main findings

In analogy to the chapters of this thesis, all important results are being summed up within this section, following the chronological order of the investigations concerned with speech rhythm in PWS and a fluently speaking control group.

6.1.1 Differences in basic speech rhythm

When analyzing speech rhythm, speech or articulation rates do not necessarily come to mind as they do not have an obvious link to rhythm like tasks of sensorimotor synchronization. However, speech and articulation rate are important global measures reflecting verbal output in a rather natural setting (Pellowski, 2010) as opposed to tasks investigating synchronization abilities that are conceptualized and constructed to investigate certain aspects of speech production under specific conditions. In speech, those rhythmically reoccurring regularities sometimes are harder to perceive due to a higher variability caused by quasi-periodic intervals. However, spoken language does indeed entail rhythm: this rhythm is carried by the amplitude envelope, as explained earlier (see chapter 2.6.1). Comparable to musical rhythm, speech shows groupings of weak and strong events that derive from metrical structures: the pattern of unstressed and stressed syllables. As discussed before, those patterns play a crucial role in speech processing and even in early language acquisition (Ladány et al., 2020). Prior to analyzing rhythm in a very specific context, it is relevant to also look at a more natural setting to analyze rhythm in rather common situations.

The first main finding of this thesis is the difference in speech and articulation rates in PWS compared to PWNS. Starting with the investigation of speech and articulation rates in two distinct settings, namely speaking and reading, it became clear that – besides the expectable result of differing speech rates – articulation rates differed significantly, too. This fact was surprising, since all instances of disfluencies had been removed in the articulation rate prior to further analyses. Rates varied significantly in both scenarios – speaking and reading. Stuttering severity showed a direct impact on perceptually fluent articulation rates in PWS, leading to a lower rate compared to PWNS – a finding that was supported by correlations of stuttering severity and the articulation rate. Not only the speech rate, but also the articulation rate displayed a higher variability, demonstrating the strong influence of stuttering – even on perceptually fluent parts of speech that might seem unimpaired at first sight.

The factor age had an impact on both speech and articulation rate as significant correlations had demonstrated. However, this fact seemed to apply rather to PWNS than to PWS. In PWS, the rise of rates usually expected with age was not as strong as within PWNS, probably due to the severity of symptoms that superimposed this effect. Effects of age are therefore rather seen as tendencies instead of clear effects.

In sum, it can be stated that speech in PWS obviously is not comparable to PWNS – even if only perceptually fluent parts are compared. Despite the absence of stuttering symptoms, the verbal output of PWS differs in many subtle ways.

6.1.2 Differences in sensorimotor synchronization

Based on the results for speech and articulation rates, a more specific approach for the investigation of verbal synchronization abilities was chosen. Here, the role of rhythm is distinctly recognizable, as the chosen paradigm involved speaking to a given rhythm. Since rhythmic processes active during speech production are still under investigation with unresolved questions – especially within the subgroup of persons affected by stuttering (Ravignani et al., 2017), gaining a better insight into these processes is highly relevant. It is hypothesized that a general internal timing deficit underlies stuttering since people who stutter show altered patterns of verbal and non-verbal synchronization abilities that can mostly be described as a less accurate, more variable rhythmic performance (Etchell et al., 2014; Falk et al., 2015; Ning et al., 2017; Olander et al., 2010; Sares et al., 2019). Especially within young PWS, research still needs to investigate this field, which is why the thesis at hand tried to add new insights into this complex topic of verbal timing deficits in PWS.

The second main finding of this thesis is the difference found between PWNS and PWS regarding their accuracy in a verbal synchronization task. As results have shown, PWS are significantly delayed in their synchronization to the beat not only when it comes to the vowel-synchronization but the onset is delayed, as well. Therefore, the issue does not seem to be the transition from consonant to vowel according to this data. Instead, results suggest a timing delay in PWS, which can be caused by at least two temporal processes taking place: it could either be altered temporal predictions that may lead to delayed temporal targets during speech production or unreliable timing mechanisms generating delays in the following process of activating syllable motor programs in PWS. Whereas the first explanation of altered temporal predictions is similar to the idea presented in the model by Harrington (1988), the second explanation is based on findings by Civier et al., (2013), who tested their hypothesis by computing a model of stuttering which involved the processes of syllable selection and initiation. Besides the fact that both reasonable explanations support the idea of altered timing in PWS leading to a delay in production, both options are in need of further investigations to actually come to a definite conclusion.

Age did not seem to play a relevant role for these investigations, as there was no significant effect of age group to be reported. In line with studies on SMS showing that children from six years on already display skills comparable to those of adults (McAuley, Jones, Holub, Johnston, & Miller, 2006), the present investigation could not report any significant changes taking place within the age-span investigated.

For the mean IVI in the unpaced condition, no significant effects of group, age group or stimulus material are to be reported, indicating a comparable performance of PWS and PWNS. The preferred mean tempo of this task hence is similar between groups and did not show any signs of aberrant processes in PWS. Results of the articulatory variability in the unpaced condition also did not display any differences between groups or age groups. However, words as the more complex stimuli showed a higher variability as opposed to syllables in both groups. Comparing variability between the unpaced and the paced condition, there is a clear effect of reduced variability observable as the metronome is added to the tasks, however, this effect of reducing variability was stronger in PWNS as opposed to PWS.

These observations lead to the assumption that it is the presence of a specific pacing event, as in this case, a metronome, that results in the strongest clearly measurable differences of speech production in PWS as opposed to PWNS. Because when looking at the paced condition, results clearly indicated a delayed synchronization pattern as described above. While altered timing therefore is the key when it comes to responding to these observations, this paradigm cannot distinguish between the different possible origins of this altered timing. Is it originated by motor or predictive timing causes? A further attempt to answer this question is made in the following paragraph.

6.1.3 Slower motor execution as a contributing factor for altered timing?

Besides this important and central finding of a different synchronization pattern that can be described as a delay in verbal synchronization, further interesting results of chapter 3 and 4 are put into relation to each other and summarized: although the comparison of duration times between groups was only assumed to be a pre-analysis to the main analyses consisting of ANOVAS to carve out group differences, the fact that already mere duration times for word and vowel productions differed significantly between groups in the unpaced and in the paced condition suggest differing processes of speech production and therefore warrant a closer look. Even without the context of external stimuli such as the metronome production times for words/syllables and vowels within them were longer in PWS as opposed to PWNS. Longer duration times – independently of contextual factors (i.e. syllabic stress, syllabic complexity, paced versus unpaced) – are to be interpreted as signs of speech systems still in development or struggling with the task of speech production and have been demonstrated before in the population of PWS in perceptive and productive studies (Colcord & Adams, 1979; Schwartze & Kotz, 2020). However, mere duration times have been sparsely investigated so far in simple speaking tasks but rather in contexts of other research questions, such as speaking rate or effects of singing (Colcord & Adams, 1979; Pellowski, 2010) and, therefore, data on this issue are hard to find.

As questions about the origin of the timing delay found cannot be answered conclusively without further investigations, this significant effect of longer durations indicates an involvement of motor components as well because longer production times for verbal output (without any external pacing) were often demonstrated in immature or compromised speech systems and also found within the first study of this thesis concerned with articulation rates (Erdemir et al., 2018; Pellowski, 2010). Combining the results of chapters 3 and 4, namely significant differences between PWS and PWNS regarding the articulation rate and mean duration times in the unpaced and paced speaking of syllables/reading of words, this raises the question whether those lower rates are to be explained by longer duration times of syllables/words and the vowels within those. The question as to what exactly caused the significant differences in the articulation rate between PWS and PWNS – as all instances of disfluencies had been removed – can potentially be answered by the duration times, that were found to differ between groups in chapter 4. Longer durations could lead to a lower rate and the mere duration was not analyzed during the process of analyzing rates – therefore, this hypothesis is merely speculative. However, while this thesis can unfortunately not determine the origin of the observed altered timing, as there were no strictly perceptive tests involved, the findings strongly suggest that there seems at least an involvement of motor components. This conclusion is based on the following observations: a lower articulation rate in reading and speaking combined with longer duration times – even in the unpaced task of speaking syllables and reading words – could indicate that PWS are confronted with a compromised speech system that needs extra time to generate verbal output leading to a delay in the activation of syllable motor programs during articulation, even when perceptually fluent (Civier et al., 2013; Olander et al., 2010). This idea does, of course, not mean that there are no perceptual components added in terms of altered temporal predictions – since the synchronization differed significantly between groups as well – but these findings can be seen as an indicator for the involvement of speech motor components resulting in altered timing.

6.1.4 Temporal parameters predict therapeutic outcome

The third main and very relevant finding of this thesis derived from the significant differences found during the investigations of chapters 3 and 4. As two quite distinct skills that both share the common ground of speech rhythm were found to vary greatly between groups, the question arises as to how this knowledge might not only serve in terms of theoretical models of speech production but on top of that how this valuable information can be used wisely in terms of improvement for diagnostics and therapy in stuttering. Research on the factors influencing persistency versus recovery in stuttering has been trying for many years, even decades, to figure out which factors are crucial for the further development of stuttering in a child affected by this disorder. Besides known factors such as age at onset or gender, most studies concerned with this question identified initial stuttering severity as an additional factor or even as the only significant predictor (Cook et al., 2013; Howell & Davis, 2011a). However, it would be a great benefit to find further, specific predictors for therapy outcome as to date, there is only little "[…] information available about variables that might predict treatment outcome in terms of post-treatment stuttering severity […]" so far (Park et al., 2021, p. 103).

Combining the knowledge about atypical rhythm being a risk factor for language disorders such as stuttering (Ladány et al., 2020), results of rhythmical differences found in PWS and the need for specific predictors, this leads to the investigation of temporal parameters, namely the articulation rate and accuracy in verbal synchronization, as potential predictors for therapeutic outcome. Since no such investigation has been done to date, it seemed reasonable to explore new paths.

Results demonstrated that rhythm, indeed, has the power to add valuable information to the question of therapeutic outcome: using linear and logistic regression analysis, chapter 5 hinted towards the fact that the articulation rate in particular is a very relevant measure as it was the only predictor that became significant in both types of analyses. Mean accuracy, however, did not reach significance as a potential predictor for therapeutic outcome.

The first analysis conducted was the linear regression to predict SSI-3 values after 15 days of intensive stuttering therapy. Here, only the articulation rate was a significant predictor as SSI-3 values pre-therapy, OASES values pre-therapy and mean accuracy did not reach significance. The second analysis conducted was a logistic regression to look at therapy outcome in a more holistic way: not only SSI-3 values but also OASES values from post-therapy were used to calculate the dichotomous outcome event of good therapy outcome versus non-respondence to therapy. In terms of logistic regression, however, not only the articulation rate, but also OASES values pre-therapy proved as significant predictors.

It can, therefore, clearly be stated that the articulation rate has the power to uncover new and relevant information regarding the question of therapeutic outcome at quite a low effort and in addition to the established diagnostic tools from the range of stuttering-specific diagnostics. As the process of generating the articulation rate from spoken and/or read speech is a lot easier than other measures of rhythmical skills or SMS and also easier to administer than further investigations of language (i.e. type-token ratio, see Cook et al., 2013) or child and parental questionnaires (Park et al., 2021), it seems reasonable to continue to use this knowledge about the potential power of the articulation rate in prognostic statements about possible therapeutic outcome.

6.2 Summary of results according to current models of speech production

A detailed summary of all results at once is not possible since hypotheses and results are too complex and multi-layered. Hence, a specific interpretation of results was given in each of the chapters. However, a recapitulatory interpretation of the overall main findings to explain speech production under the conditions of the studies in this thesis will be given here. Two models have been introduced within the theoretical background of this thesis; they will be discussed with specific regard to the results found in the three studies.

As the results of this thesis cannot conclusively differentiate between the underlying causes in terms of perceptive or productive origin, since the paradigms did not allow for a strictly separate investigation of those components, indications for both underlying causes have been found, hinting towards a combination of those processes. This seems in line with the state of the art regarding current models proposing that the generation of speech fluency demands ongoing dynamic interaction between auditory, somatosensory and speech motor networks. The dual stream model (Hickok & Poeppel, 2007) and the GODIVA-model (Civier et al., 2013; Civier et al., 2010; Guenther, 2016; Tourville & Guenther, 2011) suppose a disrupted processing of internal feedforward mechanisms and auditory feedback mechanisms along with an impaired integration of one’s own perceived speech in the speech motor planning and execution. Hence, they also incorporate a perceptive and a productive component within those models, as results of the thesis at hand indicate, too. Whereas results regarding the longer duration times and the lower articulation rate rather hint to a motor component involved, results of the synchronization task indicate that perceptive processes seem impaired within the perceptually fluent speech production in PWS.

These models of stuttering assume a disturbed motor control (Civier et al., 2013; Civier et al., 2010; Max et al., 2004) and suppose a problem in the execution of highly complex and automatized sequences of motor commands as well as a problem of vulnerability within the sensory-motor system, which seems reasonable with regard to the tasks under investigation demanding an extremely high tempo for fluent speech planning and production.

Researchers, especially in the circles around Max and colleagues (2004) and Civier and colleagues (2010, 2013), presume that PWS speak disfluently due to a shift of feedforward to feedback control during speech motor processes in combination with an exaggerated dependency on a slow and disturbed auditory feedback control system. These aberrant processes are likely to lead to mistakes in speech production, that can end in a reset of the speech motor system by repeating syllables or at least result in a slower speech production and a delayed and hence inaccurate timing (Civier et al., 2010).

6.3 Relevance of recent findings for stuttering diagnostics and therapy

A link between music and speech goes back several hundred years with music having been a an important component of treatment in various disorders of speech, such as stuttering (Jones, 2015; Ravignani et al., 2017). Variations of rhythmic speech, such as singing in particular, have gained ever-increasing interest in the clinical domain as they have shown to improve wellbeing of persons affected by various neurological diseases like dementia and Parkinson’s disease (Altenmüller & Schlaug, 2015; Osman, Tischler, & Schneider, 2016; Wan, Rübner, Hohmann, & Schlaug, 2010). Moreover, intonation-based therapy has been found to have positive effects for individuals suffering from language and speech production disorders, such as non-fluent aphasia (Schlaug, Marchina, & Norton, 2009). German speaking children displaying a specific language impairment can be treated with a therapy based on the training of word rhythm and prosodic structure to facilitate the learning of grammatical rules such as the plural and the use of articles (Penner, Fischer, & Krüge, 2006). Speaking and singing are closely linked skills, both involving rhythm as a basic competence and sharing neural correlates (Özdemir, Norton, & Schlaug, 2006). Besides the neurological evidence, different hypotheses have tried to explain those beneficial effects of singing and rhythm in various language disorders. They all share the idea of an enhanced motor control leading to an ease of symptoms either through altered patterns of articulation with special emphasis on altered prosody and rhythmic structure or socio-emotional factors promoting “[…] social connectedness, while easing the burden of communication […]” (Falk, Schreier, & Russo, 2020, p. 50).

Whereas the second explanation on social connectedness does not seem relevant for the questions at hand, as there is no singing involved, the first hypotheses of altered patterns of articulation, or to be exact an altered prosodic and rhythmic structure leading to an enhanced motor control, seems a very reasonable explanation as to why rhythmic elements/temporal parameters are worth considering as diagnostic tools (see chapters 3 and 5; articulation rate as a very basic form of rhythm with prognostic power in therapy outcome prediction) or as a therapeutic element leading to a reduction in articulatory variability as demonstrated in chapter 4. As the studies conducted in this thesis have demonstrated, there are significant differences within the perceptually fluent parts of speech of PWS that have been, to date, explained by aberrations in processes of temporal processing and the production of motor output, in this case, speech (Max et al., 2004; Wieland et al., 2015). Deficits in rhythm and timing and in skills of SMS that have been frequently observed within the population of PWS; therefore, they may as well be diagnosed and treated with rhythmical elements complementing established and evaluated stuttering therapies.

Diagnosing stuttering can be challenging as symptoms can vary considerably within the same person and as there are many aspects to be incorporated (see chapter 2.4.1). Rhythm or temporal parameters cannot add relevant information in terms of subjective or objective stuttering diagnosis. Rhythm can, however, serve as a tool for optimizing predictions about therapeutic outcome, as chapter 5 has shown, or it may even be useful as a screening tool for identifying children at risk for developing stuttering or for persisting to stutter, as Ladány et al. (2020) envisioned. They suggested a risk factor model based on a large body of evidence found to encourage this hypothesis; this risk factor model might "[…] predict the risk of developing speech/language disorders […]" (Ladány et al., 2020, p. 1) such as stuttering.

The hypothesis of CWS having problems with internal rhythm generation, as McAuley and his colleagues suggested according to their research (Wieland et al., 2015), is crucial in the case of CWS because one's ability to speak fluently is based on the competence of keeping a rhythm (Norton, 2015). It therefore seems only logical to incorporate rhythm into stuttering therapy, as it was found to be one of the core problems of this disorder. In stuttering therapy, however, rhythm is already more incorporated than one would expect: both of the two established therapeutic techniques, stuttering modification and fluency shaping (see chapter 2.4.4), already use the so called *prolonged speech* (Ingham, 1984) as a major component. It includes the prolongation of vowels to facilitate a continuous phonation; thereby, a reduction of disfluencies is achieved. This therapeutic feature, leading to a different rhythmic structuring of speech, also plays a role in singing and, therefore, some elements of rhythm have already been incorporated in the therapy of stuttering (Falk et al., 2020).

Further, more specific attempts to integrate rhythm as a major element into stuttering therapy were already made many years ago (Coppola & Yairi, 1982): metronome-paced speech training was evaluated in preschool-aged children showing positive results in terms of improvement in fluency by speaking rhythmically. While results cannot be generalized due to the small size of the sample, they demonstrated that it might be worth expanding those investigations. Many studies investigated syllable-timed speech as a form of rhythmic entrainment, showing good results in terms of a reduction of symptoms even within the population of (pre)school-aged children (L. Brown et al., 2022; Trajkovski et al., 2009). The critical issue, however, is the transfer of those rhythmical techniques into a natural way of speaking that can be used every day. For young children, in particular, but also for PWS of any age, this is the most difficult but also most important step: the transfer of learned techniques into their every-day life (Guitar, 2014; Ingham, 1984).

As rhythmic elements in stuttering therapy can be quite difficult to learn and to transfer into spontaneous speech, therapeutic singing could be a helpful tool. It might provide an additional tool for integrating rhythmic elements into established therapy, particularly for children and adolescents, as singing does not only reach physical and emotional functions but also makes therapy livelier and more enjoyable. Singing has the power to promote an environment of joy and relaxation – elements that are crucial for successful therapy, not only in children (Guitar, 2014; Thum & Mayer, 2014). It can generate an environment that strengthens the confidence in one's own speech and furthermore, it regularizes processes of breathing and vocalizing as well as coordination (Bullack, Cass, Nater, & Kreutz, 2018). A further positive effect of singing, which is of high relevance for therapeutic success is the motivation for and the feelings towards a therapy. Techniques can be learned and used, but a real success is only possible if PWS believe in themselves and are motivated to use their therapeutic tools – singing as the most social form of rhythm can promote those feelings (Altenmüller & Schlaug, 2015).

6.4 Future investigations

As the previous chapters have shown, there is a vital need for further investigations: not only in terms of research on causes and the specific role of rhythm in the emergence and the persistence of stuttering, but also in terms of rhythm and its role in stuttering diagnosis and therapy.

Even though the field of stuttering research is focusing on the role of rhythm or temporal parameters in this disorder, it has already produced a respectable number of results; there are still some questions to be answered, especially in populations that are usually underrepresented in research, such as children and adolescents. Rhythm, its neural correlates and the behavioral methods of doing research offer a wide range of questions to be raised and investigated; and although there have been many studies in the last years trying to cover all those aspects of rhythm and stuttering, a lot of questions have not been answered. The thesis at hand tried to add new information to shed more light on existing uncertainties. Indeed, it produced new results and interesting findings. However, this thesis also revealed two aspects that need to be addressed in further research.

The first is a clear consequence of the investigations on temporal parameters and SMS in PWS: for future research, it would be desirable to gain more insight into those processes of SMS in PWS of younger age with paradigms that allow for the distinction between motor or predictive timing origin. This would involve purely perceptive tests as well in order to better distinguish between the origin of found differences. This has been done before, for example by Wieland et al. (2015), who reported the rhythm perception to be impaired in CWS, and Bakhtiar, Zhang, and Sze Ki (2019), who found the processing speed to be impaired in CWS. As both studies uncovered results supporting the idea of underlying perceptive aberrations, it would be worth further investigating those issues, with a paradigm that tests comparable skills (such as verbal versus non-verbal) in both domains – perception and production. A precise differentiation between the two potential causes is not possible for the thesis at hand. This is because the testing paradigm does not allow to test separately for either the motor or the predictive timing component.

The second aspect is a result of chapter 5, which tried to use the outcome from the two previous chapters in a rather investigative way. Predicting therapeutic outcome is highly relevant, as explained before. However, research has failed to date to find further specific predictors for therapeutic outcome besides initial stuttering severity. Predictors furthermore must meet certain criteria, as they should not be too encompassing themselves. Therefore, it is a question of future research to find those predictors that fit those criteria of specificity and effectiveness. Two indications were found in the thesis at hand, namely the articulation rate and the subjective influence of stuttering on a person's life, as assessed with the OASES (Yaruss & Quesal, 2008). As this study consisted of only 39 participants, it would be worth re-testing this issue with a larger number of participants to verify the effects. Furthermore, it also needs to be determined how a temporal parameter like the articulation rate is measured most effectively during the process of pre-therapy diagnostics, as those instruments must be easily feasible by any therapist in order to make an actual difference for the standard diagnostic procedure (Guitar, 2014; Sandrieser & Schneider, 2008).

6.5 Conclusion

The goal of this thesis was to further investigate the mechanisms underlying speech production in PWS with the help of temporal parameters and rhythmical skills. Of course, it was the primary focus to add new information to this relevant topic, especially for an underrepresented group in research – children and adolescents. On top of that and besides the mere causal aspects to this research question, a first attempt to make use of this information for therapeutic purposes was started: combining the results of the first two studies on temporal parameters and rhythmical abilities in young PWS to find out whether they have the prognostic power to predict therapeutic outcome after an intensive therapy course was a quite explorative but worthwhile approach.

While the two chapters concerned with finding similarities and differences in the temporal parameters and rhythmical performances of PWS in verbal-productive tasks were able to demonstrate powerfully that even the perceptually fluent speech of PWS displays significant differences in terms of lower rates and a less accurate performance as opposed to PWNS, the chapter concerned with therapeutic outcome benefitted from parts of these findings for a differentiation between good therapy outcome and non-respondence to therapy as well as for the prediction of stuttering severity after therapy.

Whereas the latter is quite new and promising information that, once again, demonstrated the power that lies within rhythm as an indicator for so many cognitive skills (Ladány et al., 2020), the first aligns with results of previous studies that hint towards certain mechanisms but cannot fully explain the deficits underlying stuttering. However, a comprehensive explanation of this complex issue was neither a declared nor feasible goal of this thesis. The question whether stuttering speakers have a general deficit in the rhythmical patterning of speech and whether the audible and visible symptoms of stuttering are a result of this exact problem, can still be answered clearly: results of this thesis confirm the existence of a verbal-productive deficit underlying stuttering in children and adolescents who stutter. Performance in terms of the articulation rate and in verbal synchronization differed significantly from fluently speaking peers, indicating that there is an altered timing at the bottom of the stuttering phenomenon. Although the question of the origin – motor components versus predictive timing – cannot be answered conclusively and must be addressed in future research, the tendencies show that a motor component seems plausible but is probably not the sole factor in this complex construct. It rather seems that those unreliable timing mechanisms generating delays in the motor execution of verbal output also stem from altered temporal predictions, as an external pacing led to a more accurate but still significantly differing performance in PWS. It can, however, clearly be stated that speech seems to work in a different way due to different underlying timing mechanisms in stuttering speakers compared to fluently speaking persons (Etchell et al., 2015; Falk et al., 2015; Max & Yudmann, 2003; Olander et al., 2010).

Deutsche Zusammenfassung

Diese Dissertation untersucht (Sprach-)Rhythmus und dessen Relevanz für das Auftreten, die Diagnostik und die Therapie des Stotterns bei betroffenen Kindern und Jugendlichen. Ausgehend von verschiedenen Studien, die bereits Hinweise darauf geliefert haben, dass dem idiopathischen Stottern ein Rhythmus- oder Timing-Defizit zugrunde liegt (Falk et al., 2015; Olander et al., 2010; Wieland et al., 2015), untersucht auch die vorliegende Arbeit in 3 Hauptexperimenten unterschiedliche Domänen von Rhythmus in der Sprache von Kindern und Jugendlichen, die stottern. Ziel der Arbeit ist nicht nur, Ergebnisse für eine eher unterrepräsentierte Gruppe im Rahmen der Forschung zu Stottern und Rhythmus, nämlich Kinder und Jugendliche, zu liefern, sondern darüber hinaus einen präziseren Einblick in die Störungsmechanismen des Stotterns zu bekommen und herauszufinden, ob gewisse rhythmische Kompetenzen oder Profile in der Lage sind, den weiteren Verlauf der Redeflussstörung bzw. den Effekt einer Therapie vorherzusagen. In Anlehnung an Forschungsergebnisse, die Rhythmus als eine Art Screening für etwaige, in der Entwicklung auftretende Störungen untersucht haben (Ladány, et al., 2020), versucht die vorliegende Dissertation rhythmische Parameter zur Vorhersage des Therapie-Outcomes nutzbar zu machen.

Zur Untersuchung der Fragestellungen und zur Überprüfung der aufgestellten Hypothesen wurden insgesamt 54 Kinder und Jugendliche, die stottern, sowie eine Kontrollgruppe mit gleicher Alters- und Geschlechtsstruktur getestet. Während die Teilnehmer, die stottern, über eine Intensiv-Therapie (stärker-als-stottern.de) in der Nähe von München getestet wurden, fanden die Tests der Kontrollgruppen-Teilnehmer an verschiedenen Schulen in und um München statt. Aufgrund einiger Ausschlusskriterien (Komorbiditäten wie beispielsweise eine Lese-Rechtschreib-Störung oder Poltern) reduzierte sich die Anzahl der ursprünglichen Teilnehmer, eine detaillierte Beschreibung hierzu findet in den jeweiligen Kapiteln statt.

Nach einer ausführlichen theoretischen Hinführung wird im dritten Kapitel zunächst die Sprech- und Artikulationsrate bei Kindern und Jugendlichen, die stottern, sowie bei einer flüssig sprechenden Kontrollgruppe untersucht, um ein ganz grundlegendes und natürliches Maß für Rhythmus in der Sprache zu gewinnen und gleichzeitig zu untersuchen, ob und inwiefern sich der verbale Output von Teilnehmern, die stottern, unterscheidet zu dem der Kontrollgruppe. Hierzu sollten alle Teilnehmer einige Fragen zum Alltag beantworten, um spontansprachliche Äußerungen zu generieren, anschließend wurden die Teilnehmer gebeten, einen kurzen Ausschnitt aus einem Kinderbuch („Eine Woche voller Samstage“, Maar, 2008) laut vorzulesen. Somit konnte die Sprech- und Artikulationsrate in 2 Bedingungen, dem Sprechen und dem Lesen, untersucht werden. Vor allem die Artikulationsrate stellt ein sehr interessantes Maß dar, für deren Berechnung alle Symptome und Unflüssigkeiten entfernt wurden. Doch auch der Vergleich der verbleibenden, perzeptuell flüssigen Sprache brachte signifikante Unterschiede hervor und die Rate war signifikant geringer innerhalb der Kinder und Jugendlichen, die stottern. Hierbei ergaben sich auch signifikante Zusammenhänge des Stotterschweregrades mit der Sprech- und Artikulationsrate, was für die Sprechrate logisch nachvollziehbar ist. Der signifikante Zusammenhang mit der Artikulationsrate deutet darauf hin, dass auch der perzeptuell flüssige, verbale Output von Kindern und Jugendlichen, die stottern, durch die Schwere der Symptomatik beeinflusst wird und die Rate hier umso geringer wird, je stärker die Symptomatik ist. Zudem konnte auch festgestellt werden, dass das Alter positiv mit der Rate korreliert, wobei dieser Effekt stärker innerhalb der Kontrollgruppe ausgeprägt war, vermutlich aufgrund der Tatsache, dass innerhalb der Teilnehmer, die stottern, vor allem ältere Teilnehmer eine schwerere Symptomatik aufwiesen und somit der Effekt des Alters davon überlagert wurde. Schließlich konnte noch gezeigt werden, dass ein Zusammenhang zwischen den beiden Aufgaben vorhanden war, also höhere Raten beim spontanen Sprechen auch mit höheren Raten beim Lesen einhergingen.

Zusammenfassend hat diese erste Untersuchung der Sprech- und Artikulationsrate gezeigt, dass ein signifikanter Unterschied hinsichtlich des verbalen Outputs von Kindern und Jugendlichen, die stottern, gegenüber dem einer normalsprechenden Kontrollgruppe besteht. Dies betraf nicht nur die Sprechrate, sondern auch die Artikulationsrate, in jeweils beiden Modalitäten (freies Sprechen und Lesen). Die signifikanten Unterschiede hinsichtlich der Artikulationsrate deuten darauf hin, dass sich die Sprachsysteme hinsichtlich der Planung und Ausführung bedeutend zwischen den Gruppen unterscheiden: Auch der augenscheinlich symptomfreie, also flüssige Output von Kindern und Jugendlichen, die stottern, unterscheidet sich von dem der Kontrollgruppe. Dies zeigt einmal mehr, dass auch die scheinbar symptomfreien Sprachanteile und folglich auch die flüssige Sprachproduktion von Personen, die stottern, noch nicht vollständig erforscht und verstanden sind. Perzeptuell flüssige Sprache von Personen, die stottern, ist demnach also nicht direkt vergleichbar mit der von Normalsprechenden. Vielmehr deuten diese Ergebnisse darauf hin, dass sich der verbale Output von Kindern und Jugendlichen – selbst, wenn er symptomfrei ist - in vielerlei Hinsicht von dem sprachgesunder Personen unterscheidet. Diese Prozesse und die daraus resultierenden Unterschiede sind bis heute noch nicht abschließend untersucht und werden je nach Untersuchungsmethode vielleicht sogar nicht einmal erfasst. Ein Versuch, diese Details der perzeptuell flüssigen Sprache von Kindern und Jugendlichen, die stottern, zu erfassen, wurde im nächsten Abschnitt unternommen.

Eine detailliertere Analyse des flüssigen verbalen Outputs wurde daher im vierten Kapitel vorgenommen: Alle Teilnehmer wurden aufgenommen, während sie sich zu einem vorgegebenen Rhythmus synchronisierten, indem sie Silben sprachen oder Wörter vorlasen. Eine Bedingung ohne rhythmische Vorgabe diente als Vergleich beider Bedingungen (mit versus ohne rhythmische Vorgabe in Form eines Metronoms) und als Einstieg in die eigentliche Aufgabe. Dieses Vorgehen sollte einen möglichst präzisen Einblick in die sprachliche Planung und Ausführung bei Kindern und Jugendlichen, die stottern, ermöglichen. Die Ergebnisse legen nahe, dass bei der Synchronisierung zu einem vorgegebenen Rhythmus auch in den perzeptuell flüssigen Sprachanteilen signifikante Unterschiede zwischen den beiden Gruppen bestehen, die sich als eine weniger akkurate Synchronisierung der Probanden, die stottern, beschreiben lässt. Sowohl für den Vokal als auch für den Onset der Silbe/des Wortes konnte gezeigt werden, dass Kinder und Jugendliche, die stottern, signifikant später mit ihrer Synchronisierung zum Metronom beginnen, verglichen mit gleichaltrigen Kontrollprobanden. Es liegt hier also eine Verzögerung der verbalen Reaktion nicht nur in Relation zum Metronom, sondern auch im Vergleich mit normalsprechenden Kindern und Jugendlichen vor. Ein Effekt des Alters konnte hier nicht beobachtet werden. Ein Vergleich der Bedingung mit und ohne Tempovorgabe durch das Metronom konnte jedoch zeigen, dass das Metronom zu einer geringeren Variabilität der verbalen Synchronisierungsleistung führt, wobei der Effekt stärker innerhalb der Kontrollgruppe ausgeprägt war. Ein Unterschied hinsichtlich der Konsistenz der Synchronisierung zwischen den Gruppen war nicht zu beobachten. Zudem ergab die Auswertung der Bedingung ohne Tempovorgabe keine signifikanten Unterschiede zwischen den Gruppen oder Altersgruppen, sodass die Ergebnisse hier darauf hinweisen, dass vor allem die senso-motorische Synchronisation signifikante Unterschiede zwischen den Gruppen hervorbringt und auf zugrunde liegende Timing-Defizite bei Kindern und Jugendlichen, die stottern, hinweist. Es lässt sich hier demnach festhalten, dass auch perzeptuell flüssige Sprache von Kindern und Jugendlichen, die stottern deutliche Unterschiede im Vergleich zu einer flüssig sprechenden Kontrollgruppe aufweist, die als eine Verzögerung bei der motorischen Ausführung beschrieben werden kann. Es lässt sich mit diesem Testungsparadigma jedoch leider nicht eindeutig feststellen, ob die Ursache hier mehr in der Perzeption oder in der Produktion angesiedelt ist. Einen Hinweis für eine Beteiligung der produktiven Komponente liefert jedoch die Tatsache, dass sowohl in der Bedingung ohne als auch in der Bedingung mit Tempovorgabe eine signifikant längere Dauer der Vokale und der Silben/Worte innerhalb der

Gruppe der Kinder und Jugendlichen, die stottern, demonstriert wurde. Eine längere Dauer in der Vokal- oder Silben- bzw. Wortproduktion, unabhängig von der Bedingung (mit oder ohne Metronom) könnte hier ein Hinweis auf ein motorisches Defizit sein, vor allem, wenn man dies auch in Verbindung mit der Erkenntnis bringt, dass die Artikulationsrate bei Kindern und Jugendliche, die stottern, geringer, also der verbale Output motorisch verlangsamt war. Diese Ergebnisse stellen aber in keinster Weise den Ausschluss einer perzeptiven Komponente dar, denn auch die Tatsache, dass während der Bedingung mit Tempovorgabe durch das Metronom die Unterschiede zwischen den Gruppen signifikant waren hinsichtlich der Akkuratheit der Synchronisierung, sind auch wiederum ein Indiz dafür, dass die zeitliche Wahrnehmung und damit auch die angepasste Vorhersage eine Rolle für die Entstehung von Stottersymptomen spielen. Somit kann auch von einer Kombination perzeptiver und produktiver Komponenten ausgegangen werden.

Schließlich versucht das fünfte Kapitel, die Ergebnisse der vorangegangenen Untersuchungen zusammenzuführen und diese im Rahmen zweier Regressionsanalysen als mögliche Prädiktoren für das Therapie-Outcome zu nutzen. Hierfür erfolgt zunächst der Vergleich des Stotterschweregrades vor und nach der Therapie, wobei eine signifikante Verringerung der Symptomatik festgestellt werden konnte. Anschließend sollte mittels Regressionsanalyse geklärt werden, welche Faktoren diesen Erfolg am besten erklären können: Hierzu erfolgte zunächst eine lineare Regressionsanalyse, um mögliche Prädiktoren für den Stotterschweregrad nach der Therapie (gemäß SSI-3, Riley, 1994) zu ermitteln. Von den verwendeten Prädiktoren des initialen Stotterschweregrades (SSI-3), des subjektiven Einflusses des Stotterns auf den Alltag (OASES, Yaruss & Quesal, 2006), der mittleren Artikulationsrate und der mittleren Akkuratheit erwies sich lediglich die mittlere Artikulationsrate als signifikanter Prädiktor. Eine weitere Analyse, bei der mittels logistischer Regression das binäre Outcome eines guten Therapieerfolgs versus keine Veränderung durch Therapie untersucht werden sollte, ergab, dass von den gleichen Prädiktoren in diesem Fall erneut die Artikulationsrate und zusätzlich aber der initiale Wert des OASES als signifikante Prädiktoren ermittelt werden konnten. Die Ergebnisse deuten demnach darauf hin, dass eine höhere Artikulationsrate und ein geringerer initialer Schweregrad des OASES am besten das Therapie-Outcome vorhersagen können und mit einem besseren Outcome einhergehen.

Diese Ergebnisse sind als sehr explorativ anzusehen und können bislang noch nicht mit anderen Studien verglichen werden, da bisherige Untersuchungen noch nicht mit rhythmischen Parametern als Prädiktoren durchgeführt wurden. Die Ergebnisse zeigen jedoch deutlich, dass dies ein vielversprechender und interessanter Ansatz ist, der weiter untersucht werden sollte, um in der praktischen Relevanz und der konkreten therapeutischen Anwendung eine potenzielle Rolle spielen zu können.

Das Ziel dieser drei Untersuchungen war es, die genauen Mechanismen der Sprachproduktion bei Kindern und Jugendlichen, die stottern, weiter zu erforschen. Hierzu wurden vor allem zeitliche Parameter und rhythmische Kompetenzen herangezogen. Neben dem Bestreben weitere Erkenntnisse hinsichtlich der Prozesse der Sprachplanung und -Ausführung bei dieser Zielgruppe zu gewinnen, wurde darüber hinaus auch der Versuch unternommen, dieses neu gewonnene Wissen für therapeutisch-diagnostische Zwecke zu nutzen. Hierfür wurden ebendiese Erkenntnisse der Kapitel 3 und 4 in Bezug zu aktueller Literatur gesetzt, was wiederum zur Untersuchung des 5. Kapitels führte. In einer explorativen Studie wurden Ergebnisse zu den rhythmischen Leistungen der vorangegangenen Kapitel verwendet, um den Therapieerfolg vorherzusagen. Dieser Ansatz hat sich als vielversprechend herausgestellt und Potential für weitere Erforschung bewiesen.

Auch wenn die vorliegende Arbeit die Frage nach der genauen Ursache von Stottersymptomen und Abweichungen in den neurolinguistischen Systemen der Sprachverarbeitung und -Produktion bei Kindern und Jugendlichen, die stottern, nicht abschließend klären konnte, so hat diese Arbeit dennoch relevante Erkenntnisse und Zugewinne hervorgebracht. Eine finale Erklärung darüber, ob die beobachteten Timing-Defizite eher einer perzeptiven oder einer produktiven Ursache zugeschrieben werden, lässt sich hiermit zwar nicht beantworten, jedoch zeigt diese Arbeit, dass wohl eine Kombination aus beiden plausibel scheint. Eine abschließende Klärung dieser hochkomplexen Frage ist vielleicht auch nie final zu erreichen, da die zugrundeliegenden Prozesse weder durch behaviorale noch durch bildgebende Verfahren in ihrer Gänze abzubilden sind. Zusammenfassend legen die Ergebnisse dieser Dissertation nahe, dass zeitliche Parameter und sprachlicher Rhythmus für das Störungsbild Stottern eine wichtige Rolle spielen. Nicht nur in der Ursachenforschung und der modelltheoretischen Einordung der Mechanismen, die zu den Symptomen führen, ist Rhythmus von großer Bedeutung. Auch für die Diagnostik und Therapie dieses Störungsbildes birgt er ein großes Potential, das jedoch noch weiter erforscht werden muss.

References

Adams, M., & Hayden, P. (1976). The ability of stutterers and non-stutterers to initiate and terminate phonation during production of an isolated vowel. *Journal of Speech and Hearing Research, 19*(2), 290-296. doi:10.1044/jshr.1902.290

Adams, M. R., & Ramig, P. (1980). Vocal Characteristics of Normal Speakers and Stutterers during Choral Reading. *Journal of Speech and Hearing Research, 23*(2), 457-469. doi:10.1044/jshr.2302.457

Agresti, A. (2007). *An introduction to categorical data analysis*. New York: John Wiley & Sons.

Ahissar, E., Nagarajan, S., Ahissar, M., Protopapas, A., Mahncke, H., & Merzenich, M. M. (2001). Speech comprehension is correlated with temporal response patterns recorded from auditory cortex. *Proceedings of the National Academy Science of the U.S.A., 98*(23), 13367-13372. doi:10.1073/pnas.201400998

Alm, P. A. (2004). Stuttering and the basal ganglia circuits: a critical review of possible relations. *Journal of Communication Disorders, 37*, 325-369.

Alm, P. A., Karlsson, R., Sundberg, M., & Axelson, H. W. (2013). Hemispheric lateralization of motor thresholds in relation to stuttering. *PLoS ONE, 8*(10), e76824. doi:10.1371/journal.pone.0076824

Altenmüller, E., & Schlaug, G. (2015). Apollo’s gift: new aspects of neurologic music therapy. *Progress in Brain Research, 217*, 237-252. doi:10.1016/bs.pbr.2014.11.029

Ambrose, N. G., Cox, N. J., & Yairi, E. (1997). The genetic basis of persistence and recovery in stuttering. *Journal of Speech Language and Hearing Research, 40*(3), 567-580. doi:10.1044/jslhr.4003.567

Ambrose, N. G., & Yairi, E. (1999). Normative disfluency data for early childhood stuttering. *Journal of Speech Language and Hearing Research, 42*(4), 895-909. doi:10.1044/jslhr.4204.895

Ambrose, N. G., Yairi, E., Loucks, T. M., Seery, C. H., & Throneburg, R. (2015). Relation of motor, linguistic and temperament factors in epidemiologic subtypes of persistent and recovered stuttering: Initial findings. *Journal of Fluency Disorders, 45*, 12-26. doi:10.1016/j.jfludis.2015.05.004.

American-Speech-Language-Hearing-Association. (1999). Terminology pertaining to fluency and fluency disorders: guidelines. ASHA Special Interest Division 4: Fluency and Fluency Disorders. *ASHA. Supplement, 41*(2 Suppl. 19), 29-36. doi:10.1044/policy.GL1999-00063

AmericanPsychiatricAssociation. (2013). *Diagnostic and statistical manual of mental disorders* (5 ed.). Arlington, VA: American Psychiatric Publishing.

Anderson, T. K., & Felsenfeld, S. (2003). A thematic analysis of late recovery from stuttering. *American Journal of Speech-Language Pathology, 12*(2), 243-254. doi:10.1044/1058-0360(2003/070)

Andrews, G. (1985). Epidemiology of stuttering. In R. F. Curlee & W. H. Perkins (Eds.), *Nature and Treatment of Stuttering: New Directions* (pp. 1-12). San Diego: College Hill Press.

Andrews, G., & Craig, A. (1988). Prediction of Outcome After Treatment for Stuttering. *British Journal of Psychiatry, 153*, 236-240. doi:10.1192/bjp.153.2.236

Andrews, G., Craig, A., Feyer, A. M., Hoddinott, S., Howie, P., & Neilson, M. D. (1983). Stuttering: A review of research findings and theories circa 1982. *Journal of Speech and Hearing Disorders, 48*(3), 226-246. doi:10.1044/jshd.4803.226

Andrews, G., & Harris, M. (1964). *The Syndrome of Stuttering. Clinics in Developental Medicine*. London: Wilhelm Heineman Medical Books.

Andrews, G., Howie, P., Dozsa, M., & Guitar, B. (1982). Stuttering: speech pattern characteristics unter fluency-inducing conditions. *Journal of Speech and Hearing Research, 25*, 208-216.

Andrews, G., Morris-Yates, A., Howie, P., & Martin, N. (1991). Genetic factors in stuttering confirmed. *Archives of General Psychiatry, 48*(11), 1045-1035. doi:10.1001/archpsyc.1991.01810350074012

Arbisi-Kelm, T., & Jun, S. A. (2005, 10-12 September). *A comparison of disfluency patterns in normal and stuttered speech.* Paper presented at the Proceedings of Disfluency in Spontaneous Speech Workshop, Aix-en-Provence, France.

Arndt, J., & Healey, E. C. (2001). Concomitant disorders in school-age children who stutter. *Language, Speech, and Hearing Services in the Schools, 32*(2), 68-78. doi:10.1044/0161-1461(2001/006).

Aschersleben, G. (2002). Temporal control of movements in sensorimotor synchronization. *Brain and Cognition, 48*, 66-79.

Aschersleben, G. (2003). Effects of training on the timing of repetitive movements. In S. P. Shohov (Ed.), *Advances in psychology research* (pp. 15-30). Huntington, NY: Nova Science.

Backhaus, K., Erichson, B., Plinke, W., & Weiber, R. (2006). *Multivariate Analysemethoden: Eine anwendungsorientierte Einfürhung* (11th ed.). Berlin: Springer.

Bakhtiar, M., Zhang, C., & Sze Ki, S. (2019). Impaired processing speech in categorical perception: Speech perception of children who stutter. *PLoS ONE, 14*(4), e0216124. doi:10.1371/journal.pone.0216124

Beal, D. S., Cheyne, D. O., Gracco, V. L., Quraan, M. A., Taylor, M. J., & De Nil, L. F. (2010). Auditory evoked fields to vocalization during passive listening and active generation in adults who stutter. *Neuroimage, 52*(4), 1645-1653. doi:10.1016/j.neuroimage.2010.04.277

Beal, D. S., Gracco, V. L., Brettschneider, J., Kroll, R. M., & De Nil, L. F. (2013). A voxel-based morphometry (VBM) analysis of regional grey and white matter volume abnormalities within the speech production network of children who stutter. *Cortex, 49*(8), 2151-2161. doi:10.1016/j.cortex.2012.08.013

Belyk, M., Kraft, S. J., & Brown, S. (2015). Stuttering as a trait or state - an ALE meta-analysis of neuroimaging studies. *The European Journal of Neuroscience, 41*(2), 275-284. doi:10.1111/ejn.12765

Benoit, C. E., Dalla Bella, S., Farrugia, N., Obrig, H., Mainka, S., & Kotz, S. A. (2014). Musically cued gait-training improves both perceptual and motor timing in Parkinsons' Disease. *Frontiers in Human Neuroscience, 8*, 494. doi:10.3389/fnhum.2014.00494

Berens, P. (2009). CircStat: a Matlab toolbox for circular statistics. *Journal of Statistical Software, 31*, 1-21. doi:10.18637/jss.v031.i10

Bernstein Ratner, N. (1997). Linguistic behaviors at the onset of stuttering. In W. Hulstijn, H. F. M. Peters, & P. H. van Lieshout (Eds.), *Speech production: Motor control, brain research, and fluency disorders* (pp. 585-593). Amsterdam, Netherlands: Elsevier.

Biermann-Ruben, K., Samelin, R., & Schnitzler, A. (2005). Right rolandic activation during speech perception in stutterers: a MEG study. *Neuroimage, 25*(3), 793-801. doi:10.1016/j.neuroimage.2004.11.024

Birkett, E. (2014). *Experiments in time: Exploring the components of motor timing behavior in dyslexia*. Aston University, Birmingham.

Block, S., Onslow, M., Packman, A., & Dacakis, G. (2006). Connecting stuttering management and measurement: IV. Predictors of outcome for behavioral treatment for stuttering. *International Journal of Language and Communication Disorders, 41*(4), 395-406. doi:10.1080/13682820600623853

Blomgren, M. (2013). Behavioral treatments for children and adults who stutter: a review. *Psychology Research and Behavior Management, 6*, 9-19. doi:10.2147/PRBM.S31450

Blood, G. W., Ridenour Jr., V., Qualls, C. D., & Scheffner Hammer, C. (2003). Co-occuring disorders in children who stutter. *Journal of Communication Disorders, 36*(6), 427-448. doi:10.1016/s0021-9924(03)00023-6

Bloodstein, O. (1944). Studies in the psychology of stuttering: XIX. The relationship between oral reading rate and severity of stuttering. *Journal of Speech Disorders, 9*, 161-173. doi:10.1044/jshd.0902.161

Bloodstein, O. (1958). Stuttering as an anticipantory struggle reaction. In J. Eisenon (Ed.), *Stuttering: A Symposium* (pp. 1-69). New York: Harper & Brothers.

Bloodstein, O., & Bernstein Ratner, N. (2008). *A Handbook on Stuttering*. Clifton Park, NY: Delmar Learning.

Boersma, P., & Weenink, D. (2009). *Praat: doing phonetics by computer (Version 5.4.10)*. Amsterdam.

Boey, R. A., Van de Heyning, P. H., Wuyts, F. L., Heylen, L., Stoop, R., & De Bodt, M. S. (2009). Awareness and reactions of young stuttering children aged 2-7 years old towards their speech disfluency. *Journal of Communication Disorders, 42*(5), 334-346. doi:10.1016/j.jcomdis.2009.03.002

Borden, G. J. (1983). Initiation versus execution time during manual and oral counting by stutterers. *Journal of Speech and Hearing Research, 26*, 389-396.

Bosshardt, H. G. (1990). Subvocalization and reading rate differences between stuttering and nonstuttering children and adults. *Journal of Speech Language and Hearing Research, 33*(4), 776-785. doi:10.1044/jshr.3304.776

Bosshardt, H. G. (2008). *Stottern*. Göttingen: Hogrefe Verlag GmbH & Co. KG.

Bosshardt, H. G. (2010). *Frühintervention beim Stottern*. Göttingen: Hogrefe Verlag GmbH & Co. KG.

Boutsen, F. R., Brutten, G. J., & Watts, C. R. (2000). Timing and intensity variability in the metronomic speech of stuttering and nonstuttering speakers. *Journal of Speech Language and Hearing Research, 43*, 513-520.

Box, G. E. P., & Tidewell, P. W. (1962). Transformation of the Independent Variables. *Technometrics, 4*(4), 531-550. doi:10.1080/00401706.1962.10490038

Boyd, A., Dworzynski, K., & Howell, P. (2011). Pharmacological agents for developmental stuttering in children and adolescents: a systematic review. *Journal of clinical Psychopharmacology, 31*(6), 740-744. doi:10.1097/JCP.0b013e318234ee3b

Braun, A. R., Varga, M., Stager, S. V., Schulz, G., Selbie, S., Maisog, J. M., . . . Ludlow, C. L. (1997). Altered patterns of cerebral activity during speech and language production in developmental stuttering. An H2(P15)O positron emisson tomography study. *Brain, 120*(5), 761-784. doi:120/5/761/332802

Brocklehurst, P. H. (2013). Stuttering prevalence, incidence and recovery rates depend on how we define it: comment on Yairi and Ambrose' article epidemiology of stuttering: 21st century advances. *Journal of Fluency Disorders 38*, 290-293.

Brown, L., Wilson, L., Packman, A., Halaki, M., Andrews, C., O'Brian, S., . . . Menzies, R. (2022). Conversational speech of school-aged children after syllable-timed speech treatment for stuttering. *International Journal of Speech-Language Pathology, 24*(1), 42-52. doi:10.1080/17549507.2021.1946152

Brown, S., Ingham, R. J., Ingham, J. C., Laird, A. R., & Fox, P. T. (2005). Stuttered and fluent speech production: an ALE meta-analysis of functional neuroimaging studies. *Human Brain Mapping, 25*, 105-117. doi:10.1002/hbm.20140

Brysbaert, M., Buchmeier, M., Conrad, M., Jacobs, A. M., Bölte, J., & Böhl, A. (2011). The word frequency effect: A review of recent developments and implications for the choice of frequency estimates in German. *Experimental Psychology, 58*(5), 412-424. doi:10.1027/1618-3169/a000123

Buck, S., Lee, R., & Cook, F. (2002). The influence of family history of stuttering on the onset of stuttering in young children. *Folia Phoniatrica et Logopaedica, 54*(3), 117-124.

Budde, K. S., Barron, D. S., & Fox, P. T. (2014). Stuttering, induced fluency, and natural fluency: A hierarchical series of activation likelihood estimation meta-analyses. *Brain and Language, 139*, 99-107. doi:10.1016/j.bandl.2014.10.002

Bullack, A., Cass, C., Nater, U. M., & Kreutz, G. (2018). Psychobiological Effects of Choral Singing on Affective State, Social Connectedness, and Stress: Influences of Singing Activity and Time Course. *Frontiers in Behavioral Neuroscience, 12*(233). doi:10.3389/fnbeh.2018.00223

Cai, S., Tourville, J. A., Beal, D. S., Perkell, J. S., Guenther, F. H., & Ghosh, S. S. (2014). Diffusion imaging of cerebral white matter in persons who stutter: evidence for network-level anomalies. *Frontiers in Human Neuroscience, 8*, 54. doi:10.3389/fnhum.2014.00054

Canhetti de Oliveira, C. M., Broglio, G. A. F., Bernandes, A. P. L., & Capellini, S. A. (2013). Relationship between speech rate and speech disruption in cluttering. *Codas, 25*(1), 59-63. doi:10.1590/S2317-17822013000100011

Chang, S.-E., Chow, H. M., Wieland, E. A., & McAuley, J. D. (2016). Relation between functional connectivity and rhythm discrimination in children who do and do not stutter. *Neuroimage: Clinical, 25*(12), 442-450. doi:10.1016/j.nicl.2016.08.021

Chang, S.-E., Erickson, K. I., Ambrose, N. G., Hasegawa-Johnson, M. A., & Ludlow, C. L. (2008). Brain anatomy differences in childhood stuttering. *Neuroimage, 39*(3), 1333-1344. doi:10.1016/j.neuroimage.2007.09.067

Chang, S.-E., Zhu, C., Choo, A. L., & Angstadt, M. (2015). White matter neuroanatomical differences in young children who stutter. *Brain, 138*(3), 694-711. doi:10.1093/brain/awu400

Chang, S.-E., & Zhu, D. C. (2013). Neural network connectivity differences in children who stutter. *Brain 136*, 3709-3726. doi:10.1093/brain/awt275

Chen, A. H., Khalid, N. M., & Buari, N. H. (2019). Age factor affects reading acuity and reading speed in attaining text information. *International Journal of Ophtalamology, 12*(7), 1170-1176. doi:10.18240/ijo.2019.07.19

Chen, J. L., Penhune, V. B., & Zatorre, R. J. (2008). Listening to musical rhythms recruits motor regions of the brain. *Cerebral Cortex, 18*(12), 2844-2854. doi:10.1093/cercor/bhn042

Chen, J. L., Zatorre, R. J., & Penhune, V. B. (2006). Interactions between auditory and dorsal premotor cortex during synchronization to musical rhythms. *Neuroimage, 32*(4), 1771-1781. doi:10.1016/j.neuroimage.2006.04.207

Chon, H., Sawyer, J., & Ambrose, N. G. (2012). Differences of articulation rate and utterance length in fluent and disfluent utterances of preschool children who stutter. *Journal of Communication Disorders, 45*(6), 455-467. doi:10.1016/j.jcomdis.2012.08.003.

Choo, A. L., Kraft, S. J., Olivero, W., Ambrose, N. G., Sharma, H., Chang, S.-E., & Loucks, T. M. (2011). Corpus callosum differences associated with persistent stuttering in adults. *Journal of Communication Disorders, 44*(4), 470-477. doi:10.1016/j.jcomdis.2011.03.001

Civier, O., Bullock, D., Max, L., & Guenther, F. H. (2013). Computational modeling of stuttering caused by impairments in a basal ganglia thalamo-cortical-circuit involved in syllable selection and initiation. *Brain and Language, 126*, 263-278. doi:10.1016/j.bandl.2013.05.016

Civier, O., Kronfeld-Duenias, V., Amir, O., Ezrati-Vinacour, R., & Ben-Shachar, M. (2015). Reduced fractional anisotropy in the anterior corpus callosum is associated with reduced speech fluency in persistent developmental stuttering. *Brain and Language, 143*, 20-31. doi:10.1016/j.bandl.2015.01.012

Civier, O., Tasko, S. M., & Guenther, F. H. (2010). Overreliance on auditory feedback may lead to sound/syllable repetitions: simulations of stuttering and fluency-inducing conditions with a neural model of speech production. *Journal of Fluency Disorders, 35*, 246-279. doi:10.1016/j.jfludis.2010.05.002

Clark, H., & Clark, E. (1977). *Psychology and language: An introduction to psycholinguistics* New York, NY: Harcourt Brace.

Cochen De Cock, V., Dotov, D. G., Ihalainen, P., Bégel, V., F., G., Lebrun, C., . . . Dalla Bella, S. (2018). Rhythmic abilities and musical training in Parkinson’s disease: do they help? *nature partner journals, 4*(8). doi:10.1016/j.rehab.2018.05.216

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2 ed.). Hillsdale, N.J.: L. Earlbaum Associates.

Cohen, J. (2014). A new name for stuttering in DSM-5. *Monitor on Psychology, 45*(7), 51.

Colcord, R. D., & Adams, M. R. (1979). Voicing duration and vocal SPL changes associated with stuttering reduction during singing. *Journal of Speech and Hearing Research, 22*(3), 478-479.

Connally, E. L., Ward, D., Howell, P., & Watkins, K. E. (2014). Disrupted white matter in language and motor tracts in developmental stuttering. *Brain and Language, 131*, 125-135. doi:10.1016/j.bandl.2013.05.013

Conture, E. G. (1996). Treatment efficacy: stuttering. *Journal of Speech and Hearing Research, 39*(5), 18-26. doi:10.1044/jshr.3905.s18

Cook, S. (2013). Fragebogen zur psychosozialen Belastung durch das Stottern für Kinder und Jugendliche. *L.O.G.O.S. Interdisziplinär, 21*(2), 97-105.

Cook, S., Donlan, C., & Howell, P. (2013). Stuttering severity, psychosocial impact and lexical diversity as predictors of outcome for treatment of stuttering. *Journal of Fluency Disorders, 38*(2), 124-133. doi:10.1016/j.jfludis.2012.08.001

Cooper, M. H., & Allen, G. D. (1977). Timing control accuracy in normal speakers and stutterers. *Journal of Speech and Hearing Research, 20*, 55-71.

Coppola, V. A., & Yairi, E. (1982). Rhythmic speech training with preschool stuttering children: An experimental study. *Journal of Fluency Disorders, 7*(4), 447-457. doi:10.1016/0094-730X(82)90020-1

Craig, A., & Hancock, K. (1995). Self-Reported Factors Related to Relapse following Treatment for Stuttering. *Australian Journal of Human Communication Disorders, 23*(1), 48-60. doi:10.3109/asl2.1995.23.issue-1.04

Craig, A., & Tran, Y. (2005). The epidemiology of stuttering: The need for reliable estimates of prevalence and anxiety levels over the lifespan. *Advances in Speech Language Pathology, 7*(1), 41-46. doi:10.1080/14417040500055060

, F., & Port, R. F. (1998). Rhythmic constraints on stress timing in English. *Journal of Phonetics, 26*(2), 145-171. doi:10.1006/jpho.1998.0070

Cykowski, M. D., Fox, P. T., Ingham, R. J., Ingham, J. C., & Robin, D. A. (2010). A study of the reproducibility and etiology of diffusion anisotropy differences in developmental stuttering: A potential role for impaired myelination. *Neuroimage, 52*(4), 1495-1504. doi:10.1016/j.neuroimage.2010.05.011

Cykowski, M. D., Kochunov, P. V., Ingham, R. J., Ingham, J. C., Mangin, J. F., Rivière, D., . . . Fox, P. T. (2008). Perisylvian sulcal morphology and cerbral asymmetry patterns in adults who stutter. *Cerebral Cortex, 18*(3), 571-583. doi:10.1093/cercor/bhm093

Davidow, J. H. (2014). Systematic studies of modified vocalization: The effect of speech rate on speech production measures during metronome-paced speech in persons who stutter. *International Journal of Language and Communication Disorders, 49*(1), 100-112. doi:10.1111/1460-6984.12050

De Bruyn, L., Leman, M., Moelants, D., Demey, M., & Desmet, F. (2008). *Measuring and quantifying the impact of social interaction on listeners' movement to music*. Paper presented at the Proceedings of the 2008 Computers in Music Modeling and Retrieval and Network for Cross-Disciplinary Studies of Music and Meaning Conference.

De Nil, L. F., & Kroll, R. M. (2001). Searching for the neural basis of stuttering treatment outcome: recent neuroimaging studies. *Clinical Linguistigs & Phonetics, 15*(1-2), 163-168. doi:10.3109/02699200109167650

De Nil, L. F., Kroll, R. M., Kapur, S., & Houle, S. (2000). A positron emission tomography study of silent and oral single word reading in stuttering and nonstuttering adults. *Journal of Speech Language and Hearing Research, 43*(4), 1038-1053. doi:10.1044/jslhr.4304.1038

De Nil, L. F., Kroll, R. M., Lafaille, S. J., & Houle, S. (2003). A positron emission tomography study of short- and long-term treatment effects on functional brain activation in adults who stutter. *Journal of Fluency Disorders, 28*(4), 357-380. doi:10.1016/j.jfludis.2003.07.002

Dick, A. S., Bernal, B., & Tremblay, P. (2014). The language connectome: new pathways, new concepts. *Neuroscientist, 20*(5), 453-467. doi:10.1177/1073858413513502

Dworzynski, K., Remington, A., Rijsdijk, F., Howell, P., & Plomin, R. (2007). Genetic etiology in cases of recovered and persistent stuttering in an unselected, longitudinal sample of young twins. *American Journal of Speech-Language Pathology, 16*(2), 169-178. doi:10.1044/1058-0360(2007/021)

Erdemir, A., Walden, T. A., Jefferson, C. M., Choi, D., & Jones, R. M. (2018). The effect of emotion on articulation rate in persistence and recovery of childhood stuttering. *Journal of Fluency Disorders, 56*, 1-17. doi:10.1016/j.jfludis.2017.11.003

Etchell, A. C., Civier, O., Ballard, K. J., & Sowman, P. F. (2018). A systematic literature review of neuroimaging research on developmental stuttering between 1995 and 2016. *Journal of Fluency Disorders, 55*, 6-45. doi:10.1016/j.jfludis.2017.03.007

Etchell, A. C., Johnson, B. W., & Sowman, P. F. (2014). Behavioral and multimodal neuroimaging evidence for a deficit in brain timing networks in stuttering: a hypothesis and theory. *Frontiers in Human Neuroscience, 8*, 467. doi: 10.3389/fnhum.2014.00467

Etchell, A. C., Johnson, B. W., & Sowman, P. F. (2015). Beta oscillations, timing, and stuttering. *Frontiers in Human Neuroscience, 8*, 1036. doi:10.3389/fnhum.2014.01036

Etchell, A. C., Ryan, M., Martin, E., Johnson, B. W., & Sowman, P. F. (2016). Abnormal time course of low beta modulation in non-fluent preschool children: A magnetoencephalographic study of rhythm tracking. *Neuroimage, 125*, 953-963. doi:10.1016/j.neuroimage.2015.10.086

Euler, H. A., Gudenberg, A. W., v. Jung, K., & Neumann, K. (2009). Computergestützte Therapie bei Redeflussstörungen: Die langfristige Wirksamkeit der Kasseler Stottertherapie (KST). *Sprache Stimme Gehör, 33*, 193-201. doi:10.1055/s-0029-1242747

Euler, H. A., Lange, B. P., Schroeder, S., & Neumann, K. (2014). The effectiveness of stuttering treatments in Germany. *Journal of Fluency Disorders, 39*, 1-11. doi:10.1016/j.jfludis.2014.01.002

Fagnani, C., Fibiger, S., Skytthe, A., & Hjelmorg, J. V. (2011). Heritability and environmental effects for self-reported periods with stuttering: a twin study from Denmark. *Logopedics Phoniatrics Vocology, 36*(3), 114-120. doi:10.3109/14015439.2010.534503

Falk, S., Maslow, E., Thum, G., & Hoole, P. (2016). Temporal variability in sung productions of adolescents who stutter. *Journal of Communication Disorders, 62*, 101-114. doi:10.1016/j.jcomdis.2016.05.012

Falk, S., Müller, T., & Dalla Bella, S. (2015). Non-verbal sensorimotor timing deficits in children and adolescents who stutter. *Frontiers in Psychology, 6*, 847. doi:10.3389/fpsyg.2015.00847

Falk, S., Schreier, R., & Russo, F. (2020). Singing and Stuttering. In A. J. Cohen (Ed.), *The Routledge Compantion to Interdisciplinary Studies in Singing - Volume III: Wellbeing*. New York, NY: Routledge.

Felsenfeld, S., Kirk, K. M., Zhu, G., Statham, D. J., Neale, M. C., & Martin, N. G. (2000). A study of the genetic and environmental etiology of stuttering in a selected twin sample. *Behavior Genetics, 30*(5), 359-366. doi:10.1023/a:1002765620208

Field, A. (2009). *Discovering statistics usign SPSS* (3 ed.). London: Sage Publications.

Finn, P. (2004). Self-change from stuttering during adolescence and adulthood. In A. K. Bothe (Ed.), *Evidence-based treatment of stuttering: Empirical bases and applications* (pp. 117-136). Mahwah (NJ): Lawrence Erlbaum.

Finn, P., Howard, R., & Kubala, R. (2005). Unassisted recovery from stuttering: Self-perceptions of current speech behavior, attitudes, and feelings. *Journal of Fluency Disorders, 30*(4), 281-305. doi:10.1016/j.jfludis.2005.09.001

Fisher, N. I. (1993). *Statistical Analysis of Circular Data*. Cambridge Cambridge University Press.

Flaugnacco, E., Lopez, L., Terribili, C., Zoia, S., Buda, S., TIlli, S., . . . Schön, D. (2014). Rhythm perception and production predict reading abilities in developmental dyslexia. *Frontiers in Human Neuroscience, 8*, 392. doi:10.3389/fnhum.2014.00392

Foundas, A. L., Bollich, A. M., Corey, D. M., Hurley, M., & Heilman, K. M. (2001). Anomalous anatomy of speech-language areas in adults with persistent developmental stuttering. *Neurology, 57*(2), 207-215. doi:10.1212/wnl.57.2.207

Foundas, A. L., Bollich, A. M., Feldman, J., Corey, D. M., Hurley, M., Lemen, L. C., & Heilman, K. M. (2004). Aberrant auditory processing and atypical planum temporale in developmental stuttering. *Neurology, 63*(9), 1640-1646. doi:10.1212/01.wnl.0000142993.33158.2a

Foundas, A. L., Cindass, R. J., Mock, J. R., & Corey, D. M. (2013). Atypical caudate anatomy in children who stutter. *Perceptual and Motor Skills, 116*(2), 528-543. doi:10.2466/15.10.PMS.116.2.528-543

Fox, P. T., Ingham, R. J., Ingham, J. C., Hirsch, T. B., Downs, J. H., Martin, C., . . . Lancaster, J. L. (1996). A PET study of the neural systems of stuttering. *Nature, 382*, 158-161. doi:10.1038/382158a0

Fox, P. T., Ingham, R. J., Ingham, J. C., Zamparripa, F., Xiong, J. H., & Lancaster, J. L. (2000). Brain correlates of stuttering and syllable production. A PET performance-correlation analysis. *Brain, 123*, 1985-2004. doi:10.1093/brain/123.10.1985

Franke, M., Hoole, P., Schreier, R., & Falk, S. (2021). Reading Fluency in Children and Adolescents Who Stutter. *Brain Sciences, 11*(12), 1595. doi:10.3390/brainsci11121595

Frigerio-Domingues, C., & Drayna, D. (2017). Genetic contributions to stuttering: the current evidence. *Molecular Genetics & Genomic Medicine, 5*(2), 95-102. doi:10.1002/mgg3.276

Frigerio-Domingues, C., Gkalisiou, Z., Zezinka, A., Sainz, D., Gutierrez, J., Byrd, C. T., . . . Drayna, D. (2019). Genetic factors and therapy outcomes in persistent developmental stuttering. *Journal of Communication Disorders, 80*, 11-17. doi:10.1016/j.jcomdis.2019.03.007

Fujii, S., & Wan, C. Y. (2014). The role of rhythm in speech and language rehabilitation: the SEP hypothesis. *Frontiers in Human Neuroscience, 777*(8), 1-15. doi:10.3389/fnhum.2014.00777

Gick, B., Wilson, I., & Derrick, D. (2013). *Articulatory Phonetics*. West Sussex, UK: Wiley-Blackwell.

Giraud, A. L., Neumann, K., Bachoud-Levi, A.-C.-., von Gudenberg, A. W., Euler, H. A. L., H., & Preibisch, C. (2008). Severity of dysfluency correlates with basal ganglia activity in persistent developmental stuttering. *Brain and Language, 104*(2), 190-199. doi:10.1016/j.bandl.2007.04.005

Godai, U., Tatarelli, R., & Bonanni, G. (1976). Stuttering and tics in twins. *Acta geneticae medicae et gemellologiae, 25*, 369-375. doi:10.1017/s0001566000014446

Godde, E., Bailly, G., & Bosse, M.-L. (2022). Pausing and Breathing while Reading Aloud: Development from 2nd to 7th grade. *Reading and Writing, 35*(1), 1-27. doi:10.1007/s11145-021-10168-z

Goldiamond, I. (1965). Stuttering and fluency as manipulatable operant response classes. In L. Krasner & L. P. Ullmann (Eds.), *Research in Behavior Modification* (pp. 106-156). New York: Holt, Rinehart & Winston.

Goswami, U. (2019). Speech rhythm and language acquisition: An amplitude modulation phase hierarchy perspective. *Annals of the New York Academy of Sciences, 1453*, 67-78. doi:10.1111/nyas.14137

Grafman, J. (2000). Conceptualizing functional neuroplasticity. *Journal of Communication Disorders, 33*(4), 345-356. doi:10.1016/S0021-9924(00)00030-7

Grover, S., Verma, A. K., & Nebhinani, N. (2012). Clozapine-induced stuttering: a case report and analysis of similar case reports in the literature. *General Hospital Psychiatry 34*(6), 703.e701-703. doi:10.1016/j.genhosppsych.2012.02.010

Guenther, F. H. (2016). *Neural Control of Speech*. Cambridge, MA: MIT Press.

Guitar, B. (1976). Pretreatment Factors Associated with the Outcome of Stuttering Therapy. *Journal of Speech and Hearing Research, 19*(3), 590-600. doi:10.1044/jshr.1903.590.

Guitar, B. (2014). *Stuttering: An Integrated Approach to its Nature and Treatment* (4 ed.). Philadelphia, PA: Wolters Kluwer/Lippincott Williams & Wilkins.

Hall, K. D., Amir, O., & Yairi, E. (1999). A Longitudinal Investigation of Speaking Rate in Preschool Children Who Stutter. *Journal of Speech, Language, and Hearing Research, 42*, 1367-1377. doi:10.1044/jslhr.4206.1367.

Harrington, J. (1988). Stuttering, delayed auditory feedback, and linguistic rhythm. *Journal of Speech and Hearing Research, 31*(1), 36-47. doi:10.1044/jshr.3101.36

Heiberger, R., & Holland, B. (2015). *Statistical analysis and data display: An intermediate course with examples in R. Springer texts in statistics*. New York: Springer.

Heiss, W. D., Kessler, J., Thiel, A., Ghaemi, M., & Karbe, H. (1999). Differential capacity of left and right hemispheric areas for compensation of poststroke aphasia. *Annals of Neurology, 45*(4), 430-438. doi:10.1002/1531-8249(199904)45:4<430::aid-ana3>3.0.co;2-p

Hemmerich, W. (2019). StatistikGuru: Poweranalyse und Stichprobenberechnung für Regression. Retrieved from https://statistikguru.de/rechner/poweranalyse-regression.html (last viewed May 18, 2022)

Herndon, G. Y. (1967). A study of the time discrimination abilities of stutterers and nonstutterers. *Speech Monographs, 34*, 303-304.

Hickok, G., & Poeppel, D. (2004). Dorsal and ventral streams: a framework for understanding aspects of the functional anatomy of language. *Cognition, 92*(1-2), 67-99. doi:doi.org/10.1016/j.cognition.2003.10.011

Hickok, G., & Poeppel, D. (2007). The cortical organization of speech processing. *Nature Reviews Neuroscience, 8*(5), 393-402. doi:10.1038/nrn2113

Hilger, A. I., Zelaznik, H. N., & Smith, A. (2016). Evidence That Bimanual Motor Timing Performance Is Not a Significant Factor in Developmental Stuttering. *Journal of Speech, Language, and Hearing Research, 59*(4), 674-685. doi:10.1044/2016_JSLHR-S-15-0172

Holst, E., & Mittelstaedt, H. (1950). Das Reafferenzprinzip. *Naturwissenschaften, 37*(20), 464-476.

Howell, P. (2004). Cerebellar activity and stuttering: Comments on Max and Yudman (2003). *Journal of Speech Language and Hearing Research, 47*(1), 101-111. doi:10.1044/1092-4388(2004/009)

Howell, P. (2010). Behavioral effects arising from the neural substrates for atypical planning and execution of word production in stuttering. *Experimental Neurology, 225*(1), 55-59. doi:10.1016/j.expneurol.2010.06.012

Howell, P. (2011). Recovery from Stuttering. In M. J. Ball & J. S. Damico (Eds.), *Language and Speech Disorders Book Series*. New York/London: Psychology Press.

Howell, P. (2013). Screening school-aged children for risk of stuttering. *Journal of Fluency Disorders, 38*(2), 102-123. doi:10.1016/j.jfludis.2012.09.002

Howell, P., Au-Yeung, J., & Rustin, L. (1997). Clock and motor variances in lip-tracking: a comparison between children who stutter and those who do not. In W. Hulstijn, H. F. M. Peters, & P. H. van Lieshout (Eds.), *Speech Production: Motor Control, Brain Research and Fluency Disorders* (pp. 573-578). Amsterdam: Elsevier.

Howell, P., & Davis, S. (2011a). Predicting persistence of and recovery from stuttering by the teenage years based on information gathered at age 8 years. *Journal of Developmental and Behavioral Pediatrics, 32*(3), 196-205. doi:10.1097/DBP.0b013e31820fd4a9

Howell, P., & Davis, S. (2011b). The epidemiology of cluttering with stuttering. In D. Ward & K. Scaler-Scott (Eds.), *Cluttering: A handbook of research, intervention and education.* (pp. 69-89). East Sussex: Psychology Press.

Howell, P., Davis, S., & Williams, R. (2008). Late childhood stuttering. *Journal of Speech Language and Hearing Research, 51*(3), 669-687. doi:10.1044/1092-4388(2008/048)

Howie, P. M. (1981a). Concordance for stuttering in monozygotic and dizygotic twin pairs. *Journal of Speech Language and Hearing Research, 24*(3), 317-321. doi:10.1044/jshr.2403.317

Howie, P. M. (1981b). Intrapair similarity in frequency of disfluency in monocygotic and dizygotic twin pairs containing stutterers. *Behavior Genetics, 11*(3), 227-238. doi:10.1007/bf01065460

Hulstijn, W., Summers, J. J., van Lieshout, P. H., & Peters, H. F. M. (1992). Timing in finger tapping and speech: a comparison beteween stutterers and fluent speakers. *Human Movement Science, 11*(1-2), 113-124. doi:10.1016/0167-9457(92)90054-F

Huss, M., Verney, J. P., Fosker, T., Mead, N., & Goswami, U. (2011). Music, rhythm, rise time perception and developmental dyslexia: perception of musical meter predicts reading and phonology. *Cortex, 47*(6), 467-689. doi:10.1016/j.cortex.2010.07.010

Ingham, R. J. (1984). *Stuttering and behavior therapy: Current status and experimental foundations*. San Diego: College Hill Press.

Ingham, R. J., Finn, P., & Bothe, A. K. (2005). „Roadblocks“ revisited: Neural change, stuttering treatment, and recovery from stuttering. *Journal of Fluency Disorders, 30*(2), 91-107. doi:10.1016/j.jfludis.2005.01.004

Ingham, R. J., Fox, P. T., Ingham, J. C., Xiong, J. H., Zamparripa, F., Hardies, I. J., & Lancaster, J. L. (2004). Brain correlates of stuttering and syllable production: gender comparison and replication. *Journal of Speech Language and Hearing Research, 47*(2), 321-341. doi:10.1044/1092-4388(2004/026)

Ingham, R. J., Martin, R., Haroldson, S., Onslow, M., & Leney, M. (1985). Modification of listener judged naturalness in the speech of stutterers. *Journal of Speech and Hearing Research, 28*(4), 495-504. doi:10.1044/jshr.2804.495

Iverach, L., Jones, M., O'Brian, S., Block, S., Lincoln, M., Harrison, E., . . . Onslow, M. (2009). The relationship between mental health disorders and treatment outcomes among adults who stutter. *Journal of Fluency Disorders, 34*(1), 29-43. doi:10.1016/j.jfludis.2009.02.002

Jäncke, L., Hänggi, J., & Steinmetz, H. (2004). Morphological brain differences between adult stutterers and non-stutterers. *BMC Neurology, 10*(4), 23. doi:10.1186/1471-2377-4-23

Janssen, P., Kraaiamaat, F. W., & van der Meulen, S. (1983). Reading ability and disfluency in stuttering and nonstuttering elementary school children. *Journal of Fluency Disorders, 8*, 39-53. doi:10.1016/0094-730X(83)90020-7

Jentschke, S., Koelsch, S., Sallat, S., & Friederici, A. D. (2008). Children with Specific Language Impairment also show impairment of music-syntactic processing. *Journal of Cognitive Neurosciene, 20*(11), 1940-1951. doi:10.1162/jocn.2008.20135

Johannsen, H. S. (2009). Stottern bei Kindern. In M. Grohnfeldt (Ed.), *Lehrbuch der Sprachheilpädagogik und Logopädie - Band 2* (pp. 160-169). Stuttgart: Kohlhammer.

Johannsen, H. S., & Schulze, H. (1992). Abgrenzungsphänomene: Prävention und Prognose. In M. Grohnfeldt (Ed.), *Handbuch der Sprachtherapie. Band 5. Störungen der Redefähigkeit* (pp. 61-79). Berlin: Marhold.

Johannsen, H. S., & Schulze, H. (2001). Ätiologie und Verlaufsbedingungen des kindlichen Stotterns: Darstellung der Inhalte und Zielsetzungen einer Längsschnittstudie. *Sprache Stimme Gehör, 25*(1), 10-13. doi:10.1055/s-2001-11976

Johannsen, H. S., & Schulze, H. (2008). Stottern. In G. Esser (Ed.), *Lehrbuch der klinischen Psychologie und Psychotherapie bei Kindern und Jugendlichen* (3 ed., pp. 442-452). Stuttgart: Thieme.

Johnson, K. N., Walden, T. A., Conture, E. G., & Karrass, J. (2010). Spontaneous regulation of emotions in pre-school children who stutter: Preliminary findings. *Journal of Speech Language and Hearing Research, 53*(6), 1478-1496. doi:10.1044/1092-4388(2010/08-0150)

Jones, A. (2015). *Stuttering and Music Therapy.* (Honors Thesis). Ball State University, Muncie, Indiana.

Kandel, E. R., & O'Dell, T. J. (1992). Are adult learning mechanisms also used for development. *Science, 258*(5080), 243-245. doi:10.1126/science.1411522

Kang, C., Riazuddin, S., Mundorff, J., Krasnewich, D., Friedman, P., Mullikin, J. C., & Drayna, D. (2010). Mutations in the lysosomal enzyme-targeting pathway and persistent stuttering. *The New England Journal of Medicine, 362*(8), 677-685. doi:10.1056/NEJMoa0902630

Karlin, I. W. (1947). A psychosomatic theory of stuttering. *Journal of Speech Disorders, 12*(3), 319-322. doi:10.1044/jshd.1203.319

Kell, C. A., Neumann, K., Behrens, M., von Gudenberg, A. W., & Giraud, A. L. (2018). Speaking-related changes in cortical functional connectivity associated with assisted and spontaneous recovery from developmental stuttering. *Journal of Fluency Disorders, 55*, 135-144. doi:10.1016/j.jfludis.2017.02.001

Kell, C. A., Neumann, K., von Kriegstein, K., Posenenske, C., von Gudenberg, A. W., Euler, H. A., & Giraud, A. L. (2009). How the brain repairs stuttering. *Brain, 132*(10), 2747-2760. doi:10.1093/brain/awp185

Kelman, E., & Nicholas, A. (2008). *Practical intervention for early childhood stammering: Palin PCI approach*. Milton Keynes: Speechmark Publishing Ltd.

Kidd, K. K. (1980). Genetic models of stuttering. *Journal of Fluency Disorders, 5*(3), 187-201. doi:10.1016/0094-730X(80)90028-5

Kidd, K. K. (1981). Vertical transmission of susceptibility to stuttering with sex-modified expression. *Proceedings of the National Academy Science of the U.S.A., 78*(1), 606-610. doi:10.1073/pnas.78.1.606

Kikuchi, Y., Ogata, K., Umesaki, T., Yoshiura, T., Kenjo, M., Hirano, Y., . . . Tobimatsu, S. (2011). Spatiotemporal Signatures of an Abnormal Auditory System in Stuttering. *Neuroimage, 55*(3), 891-899. doi:10.1016/j.neuroimage.2010.12.083

Kim, Y.-S. G. (2015). Developmental, component-based model of reading fluency: An investigation of predictors of word-reading fluency, text-reading fluency, and reading comprehension. *Reading Research Quarterly, 50*(4), 459-481. doi:10.1002/rrq.107

Kisler, T., Reichel, U., & Schiel, F. (2017). Multilingual processing of speech via web services. *Computer Speech and Language, 45*, 326-347. doi:10.1016/j.csl.2017.01.005

Kloth, S. A. M., Janssen, P., Kraaiamaat, F. W., & Brutten, G. J. (1995). Speech-motor and linguistic skills of young stutterers prior to onset. *Journal of Fluency Disorders, 20*(2), 157-170. doi:10.1016/0094-730X(94)00022-L

Kloth, S. A. M., Kraaiamaat, F. W., Janssen, P., & Brutten, G. J. (1999). Persistence and remission of incipient stuttering among high-risk children. *Journal of Fluency Disorders, 24*(4), 253-265. doi:10.1016/S0094-730X(99)00016-9

Kohler, E., Keysers, C., Umiltà, M. A., Fogassi, L., Gallese, V., & Rissolatti, G. (2002). Hearing sounds, understanding actions: action representation in mirror neurons. *Science, 297*(5582), 846-848. doi:10.1126/science.1070311

Kotz, S. A., & Schwartze, M. (2010). Cortical speech processing unplugged: a timely subcortico-cortical framework. *Trends in Cognitive Science, 14*(9), 392-399. doi:10.1016/j.tics.2010.06.005

Kotz, S. A., & Schwartze, M. (2011). Differential input of the supplementary motor area to a dedicated temporal processing network: functional and clinical implications. *Frontiers in Integrative Neuroscience, 5*, 86. doi: 10.3389/fnint.2011.00086

Kraft, S. J., Below, J. E., Huff, C., & Yu, Y. B., J. (2015). *Australian cohort study of developmental stuttering: epidemiological and genetic analyses*. Paper presented at the 8th World Congress on Fluency Disorders of the International Fluency Association, Lisbon, Portugal.

Kraft, S. J., & Yairi, E. (2012). Genetic bases of stuttering: The State of the Art, 2011. *Folia Phoniatrica et Logopaedica, 64*(1), 34-47. doi:10.1159/000331073

Kronfeld-Duenais, V. (2014). *Speech and motor pathways in adult stuttering. Doctoral Dissertation*. Ramat-Gan (Israel): Bar-Ilan University.

Kronfeld-Duenias, V., Amir, O., Ezrati-Vinacour, R., Civier, O., & Ben-Shachar, M. (2016). The frontal aslant tract underlies speech fluency in persistent developmental stuttering. *Brain Structure and Function, 221*(1), 365-381. doi:10.1007/s00429-014-0912-8

Ladány, E., Persici, V., Fiveash, A., Tillmann, B., & Gordon, R., L. (2020). Is atypical rhythm a risk factor for developmental speech and language disorders? *Wiley interdisciplinary reviews. Cognitive science, 11*(4), 1-32. doi:10.1002/wcs.1528

Landerl, K., & Wimmer, H. (2008). Development of word reading fluency and spelling in a consistent orthography: An 8-year follow-up. *Journal of Educational Psychology, 100*(1), 150-161. doi:10.1037/0022-0663.100.1.150

Langevin, M., Huinck, W. J., Kully, D., Peters, H., Lomheim, H., & Tellers, M. (2006). A cross-cultural, long-term outcome evaluation of the ISTAR Comprehensive Stuttering Program across Dutch and Canadian adults who stutter. *Journal of Fluency Disorders, 31*(4), 229-256. doi:10.1016/j.jfludis.2006.06.001

Langevin, M., Kully, D., Teshima, S., Hagler, P., & Narasimha Prasad, N. G. (2010). Five-year longitudinal treatment outcomes of the ISTAR Comprehensive Stuttering Program. *Journal of Fluency Disorders, 35*(2), 123-140. doi:10.1016/j.jfludis.2010.04.002

Latterman, C. (2008). *Das Lidcombe-Programm zur Behandlung frühkindlichen Stotterns*. Neuss: Natke Verlag.

Latterman, C. (2011). Frühkindliches Stottern: Abwarten oder sofort behandeln? *Forum Logopädie, 25*(2), 2-7.

Leech, K. A., Bernstein Ratner, N., Brown, B., & Weber, C. (2017). Preliminary evidence that growth in productive language differentiates childhood stuttering persistence and recovery. *Journal of Speech, Language, and Hearing Research, 60*, 3097-3109. doi:10.1044/2017_JSLHR-S-16-0371

Lewis, A. C. (2007). *Rhythmus*. Kassel: Gustav Bosse Verlag.

Lewis, P. A., Wing, A. M., Pope, P. A., P., P., & Miall, R. C. (2004). Brain activity correlates differentially with increasing temporal complexity of rhythms during initialisation, synchronisation, and continuation phases of paced finger tapping. *Neuropsychologia, 42*(10), 1301-1312. doi:10.1016/j.neuropsychologia.2004.03.001

Logan, K. J., Byrd, C. T., Mazzocchi, M., & Gillam, R. B. (2011). Speaking rate characteristics of elementary school-aged children who do and do not stutter. *Journal of Communication Disorders, 44*(1), 130-147. doi:10.1016/j.jcomdis.2010.08.001

Loucks, T. M., Kraft, S. J., Choo, A. L., Sharma, H., & Ambrose, N. G. (2011). Functional brain activation differences in stuttering identified with a rapid fMRI sequence. *Journal of Fluency Disorders, 36*(4), 302-307. doi:10.1016/j.jfludis.2011.04.004

Lu, C., Chen, C., Peng, D., You, W., Zhang, X., Ding, G., . . . Howell, P. (2012). Neural anomaly and reorganization in speakers who stutter: a short-term intervention study. *Neurology, 79*(7), 625-632. doi:10.1212/WNL.0b013e31826356d2

Lu, C., Peng, D., Chen, C., Ning, N., Ding, G., Li, K., . . . Lin, C. (2010). Altered effective connectivity and anomalous anatomy in the basal ganglia-thalamocortical circuit of stuttering speakers. *Cortex, 46*(1), 49-67. doi:10.1016/j.cortex.2009.02.017

Lu, C., Zheng, L., Long, Y., Yan, Q., Ding, G., Liu, L., . . . Howell, P. (2017). Reorganization of brain function after a short-term behavioral intervention for stuttering. *Brain and Language, 168*, 12-22. doi:10.1016/j.bandl.2017.01.001

Ludlow, C. L., & Loucks, T. M. (2003). Stuttering: a dynamic motor control disorder. *Journal of Fluency Disorders, 28*(4), 273-295. doi:10.1016/j.jfludis.2003.07.001

Lundgren, K., Helm-Estabrooks, N., & Klein, R. (2010). Stuttering Following Acquired Brain Damage: A Review of the Literature. *Journal of Neurolinguistics, 23*(5), 447-454. doi:10.1016/j.jneuroling.2009.08.008

Maar, P. (2008). *Eine Woche voller Samstage*. München: Deutscher Taschenbuch Verlag GmbH & Co.KG.

Mahr, G., & Leith, W. (1992). Psychogenic stuttering of adult onset. *Journal of Speech and Hearing Research, 35*(2), 283-386. doi:10.1044/jshr.3502.283

Månsson, H. (2000). Childhood stuttering: Incidence and development. *Journal of Fluency Disorders, 25*, 47-57. doi:10.1016/S0094-730X(99)00023-6

Mardia, K. V., & Jupp, P. E. (2000). *Directional Statistics*. New York: John Wiley.

MATLAB. (2017). Version 9.3.0.713579 (R2017b). Natick, Massachusetts: The MathWorks Inc.

Max, L., & Gracco, V. L. (2005). Coordination of oral and laryngeal movements in the perceptually fluent speech of adults who stutter. *Journal of Speech Language and Hearing Research, 48*(3), 524-542. doi:10.1044/1092-4388(2005/036)

Max, L., Guenther, F. H., Gracco, V. L., Gosh, S., & Wallace, M. (2004). Unstable or insufficiently activated internal models and feedback-biased motor control as sources of disfluency: a theoretical model of stuttering. *Contemporary issues in communication science and disorders, 31*, 105-122.

Max, L., & Yudmann, E. A. (2003). Accuracy and variability of isochronous rhythmic timing across motor systems in stuttering versus nonstuttering indiviudals. *Journal of Speech Language and Hearing Research, 46*(1), 146-163. doi:10.1044/1092-4388(2003/012)

May, A., & Gaser, C. (2006). Magnetic resonance-based morphometry: a window into structural plasticity of the brain. *Corrent Opinion in Neurology, 19*(4), 407-411. doi:10.1097/01.wco.0000236622.91495.21

McAuley, J. D., Jones, M. R., Holub, S., Johnston, H. M., & Miller, N. S. (2006). The time of our lives: Lifespan development of timing and event tracking. *Journal of Experimental Psychology: General, 135*(3), 348-367. doi:10.1037/0096-3445.135.3.348

McClean, M. D., Kroll, R. M., & Loftus, N. S. (1990). Kinematic analysis of lip closure in stutterers' fluent speech. *Journal of Speech and Hearing Research, 33*(4), 755-760. doi:10.1044/jshr.3304.755

Metten, C., Zückner, H., & Rosenberger, S. (2007). Evaluation einer Stotterintensivtherapie mit Kindern und Jugendlichen. *Sprache Stimme Gehör, 31*(2), 72-78. doi:10.1055/s-2007-958631

Meyers, S., & Freeman, F. (1985). Mother and child speech rates as a variable of stuttering and disfluency. *Journal of Speech and Hearing Research, 28*(3), 436-444. doi:10.1044/jshr.2803.444

Misaghi, E., Zhang, Z., Gracco, V. L., De Nil, L. F., & Beal, D. S. (2018). White matter tractography of the neural network for speech-motor control in children who stutter. *Neuroscience Letters, 668*, 37-42. doi:10.1016/j.neulet.2018.01.009

Monier, F., & Droit-Volet, S. (2019). Development of sensorimotor synchronization abilities: Motor and cognitive components. *Child Neuropsyhcology, 25*(8), 1043-1062. doi:10.1080/09297049.2019.1569607

Montgomery, B. M., & Fitch, J. L. (1988). The prevalence of stuttering in the hearing-impaired school age population. *Journal of Speech and Hearing Disorders, 53*(2), 131-135. doi:10.1044/jshd.5302.131

Moore, W. H. J. (1984a). Central nervous system characteristics of stutterers. In R. F. Curlee & W. H. Perkins (Eds.), *Nature and treatment of stuttering: New directions* (pp. 49-71). San Diego (CA): College Hill Press.

Moore, W. H. J. (1984b). Hemispheric alpha asymmetries during an electromyographic biofeedback procedure for stuttering: A single-subject experimental design. *Journal of Fluency Disorders, 9*(2), 143-162. doi:10.1016/0094-730X(84)90032-9

Moore, W. H. J., & Haynes, W. O. (1980). Alpha hemispheric asymmetry and stuttering: Some support for a segmentation dysfunction hypothesis. *Journal of Speech and Hearing Research, 23*(2), 229-247. doi:10.1044/jshr.2302.229

Myers, F. L., Bakker, K., St.Louis, K. O., & Raphael, L. J. (2012). Disfluencies in cluttered speech. *Journal of Fluency Disorders, 27*(1), 9-19. doi:10.1016/j.jfludis.2011.10.001

Namasivayam, A. K., & van Lieshout, P. H. (2011). Speech motor skill and stuttering. *Journal of Motor Behavior, 43*(3), 477.489. doi:10.1080/00222895.2011.628347

Natke, U., & Alpermann, A. (2010). *Stottern - Erkenntnisse, Theorien, Behandlungsmethoden* (3 ed.). Bern: Verlag Hans Huber, Hogrefe AG.

Natke, U., Alpermann, A., Heil, W., Kuckenberg, S., & Zückner, H. (2010). Langzeiteffekte der Intensiv-Modifikation Stottern (IMS). *Sprache Stimme Gehör, 34*(3), 155-164. doi:10.1055/s-0029-1246202

Natke, U., Sandrieser, P., Pietrowsky, R., & Kalveram, K. T. (2006). Disfluency data for German preschool children who stutter and comparison children. *Journal of Fluency Disorders, 31*(3), 165-176. doi:10.1016/j.jfludis.2006.04.002

Natke, U., Sandrieser, P., van Ark, M., Pietrowsky, R., & Kalveram, K. T. (2004). Linguistic stress, within-word-position, and grammatical class in Relation to Early Childhood Stuttering. *Journal of Fluency Disorders, 29*(2), 109-122. doi:10.1016/j.jfludis.2003.11.002

Neef, N. E., Anwander, A., & Friederici, A. D. (2015). The neurobiological grounding of persistent stuttering: From structure to function. *Current Neurology and Neuroscience Reports, 15*(9), 579. doi:10.1007/s11910-015-0579-4

Neef, N. E., Jung, K., Rothkegel, H., Pollock, B., von Gudenberg, A. W., Paulus, W., & Sommer, M. (2011). Right-shift for non-speech motor processing in adults who stutter. *Cortex, 47*(8), 945-954. doi:10.1016/j.cortex.2010.06.007

Neef, N. E., Paulus, W., Neef, A., von Gudenberg, A. W., & Sommer, M. (2011). Reduced intracortical inhibition and facilitation in the primary motor tongue representation of adults who stutter. *Clinical Neurophysiology, 122*(9), 1802-1811. doi:10.1016/j.clinph.2011.02.003

Neef, N. E., Sommer, M., Neef, A., Paulus, W., von Gudenberg, A. W., Jung, K., & Wüstenberg, T. (2012). Reduced speech perceptual acuity for stop consonants in individuals who stutter. *Journal of Speech Language and Hearing Research, 55*(1), 276-289. doi:10.1044/1092-4388(2011/10-0224)

Neilson, M. D., & Neilson, P. D. (1987). Speech motor control and stuttering: a computational model of adaptive sensory-motor processing. *Speech Communication, 6*(4), 325-333. doi:10.1016/0167-6393(87)90007-0

Neumann, K. (2007). Stottern im Gehirn: neue Erkenntnisse aus Humangenetik und Neurowissenschaften. *Forum Logopädie, 2*(21), 6-13.

Neumann, K., & Euler, H. A. (2010). Neuroimaging in stuttering. In B. Guitar & R. McCauley (Eds.), *Treatment of stuttering. Established and Emerging interventions* (pp. 355-377). Baltimore (MD): Lippincott, Williams & Wilkins.

Neumann, K., Euler, H. A., Bosshardt, H. G., Cook, S., Sandrieser, P., Schneider, P., . . . Thum, G. (2016). Pathogenese, Diagnostik und Behandlung von Redeflussstörungen. Evidenz- und konsensbasierte S3- Leitlinie. from Deutsche Gesellschaft für Phoniatrie und Pädaudiologie (http://www.awmf.org/leitlinien/detail/ll/049-013.html, last viewed June 12, 2021)

Neumann, K., Euler, H. A., Bosshardt, H. G., Cook, S., Sandrieser, P., & Sommer, M. (2017). The Pathogenesis, Assessment and Treatment of Speech Fluency Disorders. *Deutsches Ärtzeblatt International, 114*, 383-390. doi:10.3238/arztebl.2017.0383

Neumann, K., Euler, H. A., von Gudenberg, A. W., Giraud, A. L., Lanfermann, H., Gall, V., & Preibisch, C. (2003). The nature and treatment of stuttering as revealed by fMRI. A within- and between-group comparison. *Journal of Fluency Disorders, 28*(4), 381-410. doi:10.1016/j.jfludis.2003.07.003

Neumann, K., Preibisch, C., Euler, H. A., von Gudenberg, A. W., Lanfermann, H., Gall, V., & Giraud, A. L. (2005). Cortical Plasticity Associated With Stuttering Therapy. *Journal of Fluency Disorders, 30*(1), 23-39. doi:10.1016/j.jfludis.2004.12.002

Ning, N., Peng, D., Liu, X., & Yang, S. (2017). Speech Timing Deficit of Stuttering: Evidence from Contingent Negative Variations. *PLoS ONE, 12(1)*(e0168836), 1-17. doi:10.1371/journal.pone.0168836

Nippold, M. A. (2012). Stuttering and language ability in children: Questioning the connection. *American Journal of Speech-Language Pathology, 21*(3), 183-196. doi:10.1044/1058-0360(2012/11-0078)

Nippold, M. A. (2018). Language development in children who stutter: A review of recent research. *International Journal of Speech-Language Pathology, 11*, 109. doi:10.1080/17549507.2018.1457721

Nippold, M. A., & Schwarz, I. E. (1990). Reading disorders in stuttering children. *Journal of Fluency Disorders, 15*(3), 175-189. doi:10.1016/0094-730X(90)90017-M

Noreika, V., Falter, C. M., & Rubia, K. (2013). Timing deficits in attention-deficit/hyperactivity disorder (ADHD): evidence from neurocognitive and neuroimaging studies. *Neuropsychologia, 51*(2), 235-266. doi:10.1016/j.neuropsychologia.2012.09.036

Norton, A. (2015). For Kids Who Stutter, Rhythm Perception May Be Key. *HealthDay Reporter.*

O'Brian, S., Cream, A., Packman, A., & Onslow, M. (2001). A replicable, nonprogrammed, instrumentfree method for the control of stuttering with prolonged speech. *Asia Pacific Journal of Speech, Language and Hearing, 6*(2), 91-96. doi:10.1179/136132801805576680

O'Brian, S., Onslow, M., Cream, A., & Packman, A. (2003). The Camperdown programm: Outcomes of a new prolonged-speech treatment model. *Journal of Speech Language and Hearing Research, 46*(4), 933-946. doi:10.1044/1092-4388(2003/073)

Ochsenkühn, C., Frauer, C., & Thiel, M. M. (2015). *Stottern bei Kindern und Jugendlichen*. Heidelberg: Springer Medizin Verlag.

Olander, L., Smith, A., & Zelaznik, H. N. (2010). Evidence that a motor timing deficit is a factor in the development of stuttering. *Journal of Speech Language and Hearing Research, 53*(4), 876-886. doi:10.1044/1092-4388(2009/09-0007)

Onslow, M., Packman, A., & Harrison, E. (2003). *The Lidcombe Program of Early Stuttering Intervention*. Austin, TX: Pro-Ed.

Ooki, S. (2005). Genetic and environmental influences on stuttering and tics in Japanese twin children. *Twin Research and Human Genetics, 8*(1), 69-75. doi:10.1375/1832427053435409

Osman, S. E., Tischler, V., & Schneider, J. (2016). 'Singing for the Brain': A qualitative study exploring the health and well-being benefits of singing for people with dementia and their carers. *Dementia (London), 15*(6), 1326-1339. doi:10.1177/1471301214556291

Özdemir, E., Norton, A., & Schlaug, G. (2006). Shared and distinct neural correlates of singing and speaking. *Neuroimage, 33*, 628-635. doi:10.1016/j.neuroimage.2006.07.013

Paden, E. P., Yairi, E., & Ambrose, N. G. (1999). Early childhood stuttering II: Initial status of phonological abilities. *Journal of Speech Language and Hearing Research, 42*(5), 1113-1124. doi:10.1044/jslhr.4205.1113

Pardoe, I. (2012). *Applied Regression Modeling: A Business Approach*: Wiley.

Park, V., Onslow, M., Lowe, R., Jones, M., O'Brian, S., Packman, A., . . . Hewat, S. (2021). Predictors of Lidcombe Program treatment dropout and outcome for early stuttering. *International Journal of Language and Communication Disorders, 56*(1), 102-115. doi:10.1111/1460-6984.12586

Parrell, B., Lammert, A. C., Ciccarelli, G., & Quatieri, T. F. (2019). Current models of speech motor control: A control-theoretic overview of architectures and properties. *The Journal of the Acoustic Society of America, 145*(3), 1456-1481. doi:10.1121/1.5092807

Pellowski, M. W. (2010). Speech-Language Pathologists' Knowledge of Speaking Rate and Its Relationship to Stuttering. *Contemporary issues in communication science and disorders, 37*, 50-57. doi:10.1044/cicsd_36_S_50

Penner, Z., Fischer, A., & Krüge, C. (2006). *Von der Silbe zum Wort. Rhythmus und Wortbildung in der Sprachförderung*. Troisdorf: Bildungsverlag EINS.

Peters, H. F. M., Hulstijn, W., & Starkweather, C. W. (1989). Acoustic and physiological reaction times of stutterers and nonstutterers. *Journal of Speech and Hearing Research, 32*(3), 668-680. doi:10.1044/jshr.3203.668

Peters, H. F. M., Hulstijn, W., & van Lieshout, P. H. (2000). Recent developments in speech motor research into stuttering. *Folia Phoniatrica et Logopaedica, 52*(1-3), 103-119. doi:10.1159/000021518

Pine, J. (2011). Book of African-American Quotations. In S. L. Rattiner (Ed.), *Dover Thrift Editions*. Mineola, New York: Dover Publications, Inc.

Pinto, J., Schiefer, A. M., & Ávila, C. (2013). Disfluency and speech rate in spontaneous production and in oral reading in people who stutter and who do not stutter. *Audiology - Communication Research, 18*(2), 63-70. doi:10.1590/S2317-64312013000200003

Pinto, J. S., Picoloto, L. A., Capellini, S. A., Palharini, T. A., & Canhetti de Oliveira, C. M. (2021). Fluency and reading comprehension in students with and without stuttering. *Codas, 33*(5), e20200059. doi:10.1590/2317-1782/20202020059

Preibisch, C., Neumann, K., Raab, P., Euler, H. A., von Gudenberg, A. W., Lanfermann, H., & Giraud, A. L. (2003). Evidence for compensation for stuttering by the right frontal operculum. *Neuroimage, 20*(2), 1356-1364. doi:10.1016/S1053-8119(03)00376-8

Proctor, A., Yairi, E., Duff, M. C., & Zhang, J. (2008). Prevalence of stuttering in African American preschoolers. *Journal of Speech Language and Hearing Research, 51*(6), 1465-1479. doi:10.1044/1092-4388(2008/07-0057)

Rasinski, T. (2004). Creating Fluent Readers: A growing body of evidence points to reading fluency as an important factor in student reading success. *Educational Leadershit, 61*(6), 46-51.

Rathcke, T., Lin, C., Falk, S., & Dalla Bella, S. (2021). Tapping into linguistic rhythm. *Laboratory Phonology, 12*(1), 11. doi:10.5334/labphon.248

Rautakoski, P., Hannus, T., Simberg, S., Sandnabba, N. K., & Santtila, P. (2012). Genetic and environmental effects on stuttering: a twin study from Finland. *Journal of Fluency Disorders, 37*(3), 202-2010. doi:10.1016/j.jfludis.2011.12.003

Ravignani, A., Honing, H., & Kotz, S. A. (2017). Editorial: The Evolution of Rhythm Cognition: Timing in Music and Speech. *Frontiers in Human Neuroscience, 11*, 303. doi:10.3389/fnhum.2017.00303

Reed, P., & Wu, Y. (2013). Logistic regression for risk factor modelling in stuttering research. *Journal of Fluency Disorders, 38*(2), 88-101. doi:10.1016/j.jfludis.2012.09.003

Reilly, S., Onslow, M., & Packman, A. (2013). Natural history of stuttering to 4 years of age: A prospective community-based Study. *Pediatrics, 132*(3), 460-467. doi:10.1542/peds.2012-3067

Reilly, S., Onslow, M., Packman, A., Wake, M., Bavin, E., & Prior, M. (2009). Predicting stuttering onset by age of 3: A prospective, community cohort study. *Pediatrics, 123*(1), 170-177. doi:10.1542/peds.2007-3219

Repp, B. H. (2005). Sensorimotor synchronization: a review of the tapping literature. *Psychonomic Bulletin and Review, 12*(6), 969-992. doi:10.3758/BF03206433

Repp, B. H. (2010). Sensorimotor synchronisation and perception of timing: Effects of music training and task experience. *Human Movement Science, 29*, 200-213. doi:10.1016/j.humov.2009.08.002

Repp, B. H., & Su, Y. H. (2013). Sensorimotor synchronization: a review of recent research (2006-2012). *Psychonomic Bulletin and Review, 20*(3), 403-452. doi:10.3758/s13423-012-0371-2.

Riley, G. D. (1994). *Stuttering Severity Instrument for Children and Adults* (3 ed.). Austin, Texas: Pro Ed.

Riley, G. D. (2009). *Stuttering Severity Instrument - Fourth Edition (SSI-4)*. Austin, Texas: Pro Ed.

Rispoli, M. (2003). Changes in the nature of sentence production during the period of grammatical development. *Journal of Speech Language and Hearing Research, 46*(818-830). doi:10.1044/1092-4388(2003/064)

Rosen, H. J., Petersen, S. E., Linenweber, M. R., Snyder, A. Z., White, D. A., Chapman, L., . . . Corbetta, M. D. (2000). Neural correlates of recovery from aphasia after damage to left inferior frontal cortex. *Neurology, 55*(12), 1883-1894. doi:10.1212/wnl.55.12.1883

Rozental, A., Andersson, G., & Carlbring, P. (2019). In the Absence of Effects: An Individual Patient Data Meta-Analysis fo Non-response and Its Predictors in Internet-Based Cognitive Behavior Therapy. *Frontiers in Psychology, 10*, 589. doi:10.3389/fpsyg.2019.00589

Sallat, S. (2011). Prosodische und musikalische Verarbeitung im gestörten Spracherwerb. *Sprache Stimme Gehör, 35*(3), 105-111. doi:10.1055/s-0031-1284407

Salmelin, R., Schnitzler, A., Schmitz, F., & Freund, H. J. (2000). Single word reading in developmental stutterers and fluent speakers. *Brain, 123*(6), 1184-1202. doi:10.1093/brain/123.6.1184

Sandrieser, P., & Schneider, P. (2008). *Stottern im Kindesalter* (3 ed.). Stuttgart: Georg Thieme Verlag.

Sares, A. G., Deroche, M. L. D., Shiller, D. M., & Gracco, V. L. (2018). Timing variability of sensorimotor integration during vocalization in individuals who stutter. *Scientific Reports, 8*, 16340. doi:10.1038/s41598-018-34517-1

Sares, A. G., Deroche, M. L. D., Shiller, D. M., & Gracco, V. L. (2019). Adults who stutter and metronome synchronization: evidence for a nonspeech timing deficit. *Annals of New York Academy of Sciences, 1449*(1), 56-59. doi:10.1111/nyas.14117

Sawyer, J., Chon, H., & Ambrose, N. G. (2008). Influence of rate, length, and complexity on speech disfluency in a single speech sample in preschool children who stutter. *Journal of Fluency Disorders, 33*(3), 220-240. doi:10.1016/j.jfludis.2008.06.003

Schiel, F. (1999). *Automatic Phonetic Transcription of Non-Prompted Speech.* Paper presented at the Proceedings of the XIVth International Congress of Phonetic Sciences : ICPhS 99, San Francisco.

Schlaug, G., Marchina, S., & Norton, A. (2009). Evidence for plasticity in white-matter tracts of patients with chronic Broca's aphasia undergoing intense intonation-based speech therapy. *Annals of New York Academy of Sciences 1169*, 385-394. doi:10.1111/j.1749-6632.2009.04587

Schwartze, M., & Kotz, S. A. (2016). Contributions of Cerebellar Event-Based Temproal Processing and Preperatory Function to Speech Perception. *Brain and Language, 161*, 28-32. doi:10.1016/j.bandl.2015.08.005

Schwartze, M., & Kotz, S. A. (2020). Decreased sensitivity to changing durational parameters of syllable sequences in people who stutter. *Language, Cognition and Neuroscience, 35*(2), 179-187. doi:10.1080/23273798.2019.1642499

Shugart, Y. Y., Mundorff, J., Kilshaw, J., Doheny, K., Doan, B., Green, D. M., & Wanyee, J. (2004). Results of a genome-wide linkage-scan for stuttering. *American Journal of Medical Genetics. Part A, 124A*(2), 133-135. doi:10.1002/ajmg.a.20347

Smith, A., Goffman, L., Sasisekaran, J., & Weber-Fox, C. (2012). Language and motor abilities of preschool children who stutter: evidence from behavioral and kinematic indices of nonword repetition performance. *Journal of Fluency Disorders, 37*(4), 344-358. doi:10.1016/j.jfludis.2012.06.001

Smith, A., Sadagopan, N., Walsh, B., & Weber-Fox, C. (2010). Increasing phonological complexity reveals heightened instability in inter-articulatory coordination in adults who stutter. *Journal of Fluency Disorders, 35*(1), 1-18. doi:10.1016/j.jfludis.2009.12.001

Smits-Bandstra, S., De Nil, L. F., & Saint-Cyr, J. A. (2006). Speech and nonspeech sequence skill learning in adults who stutter. *Journal of Fluency Disorders, 31*(2), 116-136. doi:10.1016/j.jfludis.2006.04.003

Sommer, M., & Büchel, C. (2004). What causes stuttering. *PLoS Biology, 2*(2), 0159-0163. doi:10.1371/journal.pbio.0020046

Sommer, M., Koch, M. A., Paulus, W., Weiller, C., & Büchel, C. (2002). Disconnection of speech-relevant brain areas in persistent developmental stuttering. *Lancet, 360*(9330), 380-383. doi:10.1016/S0140-6736(02)09610-1

Sommer, M., Wischer, S., Tergau, F., & Paulus, W. (2003). Normal Intracortical Excitability in Developmental Stuttering. *Movement Disorders, 18*(7), 826-830. doi:10.1002/mds.10443

Sowinski, J., & Dalla Bella, S. (2013). Poor synchronization to the beat may result from deficient auditory-motor mapping. *Neuropsychologia, 51*(10), 1952-1963. doi:10.1016/j.neuropsychologia.2013.06.027

Sowman, P. F., Crain, S., Harrison, E., & Johnson, B. W. (2014). Lateralization of brain activation in fluent and non-fluent preschool children: a magnetencephalographic study of picture-naming. *Frontiers in Human Neuroscience, 8*, 354. doi:10.3389/fnhum.2014.00354

Stager, S. V., Jeffries, K. J., & Braun, A. R. (2003). Common features of fluency-evoking conditions studied in stuttering subjects and controls: an H215O PET study. *Journal of Fluency Disorders, 28*(4), 319-336. doi:10.1016/j.jfludis.2003.08.004

Starkweather, C. W. (1980). A multiprocess behavioral approach to stuttering therapy. *Seminars in Speech, Language and Hearing, 1*(4), 327-337.

Starkweather, C. W. (1987). *Fluency and Stuttering*. Englewood Cliffs, NJ: Prentice-Hall.

Starkweather, C. W., & Gottwald, S. R. (1993). A pilot study of relations among specific measures obtained at intake and discharge in a program of prevention and early intervention for stuttering. *American Journal of Speech-Language Pathology, 2*(1), 51-58. doi:10.1044/1058-0360.0201.51

Stone, E. R. (2010). t Test, Paired Samples. In N. J. Salkind (Ed.), *Encyclopedia of research design* (pp. 1560-1565). Los Angeles: SAGE.

Suresh, R., Ambrose, N. G., Roe, C., Pluzhnikov, A., Wittke-Thompson, J. K., Ng, M. C., . . . Cox, N. J. (2006). New complexities in the genetics of stuttering: Significant sex-specific linkage signals. *American Journal of Human Genetics, 78*(4), 554-563. doi:10.1086/501370

Tabachnick, B. G., & Fidell, L. S. (2018). *Using Multivariate Statistics* (7th ed.): Pearson Education.

Thomson, J. M., Fryer, B., Maltby, J., & Goswami, U. (2006). Auditory and motor rhythm awareness in adults with dysleia. *Journal of Research in Reading, 29*(3), 334-348. doi:10.1111/j.1467-9817.2006.00312.x

Thomson, J. M., & Goswami, U. (2008). Rhythmic processing in children with developmental dyslexia: auditory and motor rhythms link to reading and spelling. *Journal of Physiology-Paris, 102*(1-3), 120-129. doi:10.1016/j.jphysparis.2008.03.007

Thum, G., & Mayer, I. (2014). *Stottertherapie bei Kindern und Jugendlichen: Ein methodenkombinierter Ansatz*. München: Ernst Reinhardt.

Tierney, A., & Kraus, N. (2013). The ability to tap to a beat relates to cognitive, linguistic, and perceptual skills. *Brain and Language, 124*(3), 225-231. doi:10.1016/j.bandl.2012.12.014

Tierney, A., & Kraus, N. (2014). Auditory-motor entrainment and phonological skills: precise auditory timing hypothesis (PATH). *Frontiers in Human Neuroscience, 8*, 1-9. doi:10.3389/fnhum.2014.00949

Tourville, J. A., & Guenther, F. H. (2011). The DIVA model: A neural theory of speech acquisition and production. *Language and Cognitive Processes, 25*(7), 952-981. doi:10.1080/01690960903498424

Toyomura, A., Fujii, T., & Kuriki, S. (2011). Effect of external auditory pacing on the neural activity of stuttering speakers. *Neuroimage, 57*(4), 1507-1516. doi:10.1016/j.neuroimage.2011.05.039

Trajkovski, N., Andrews, C., Onslow, M., Packman, A., O'Brian, S., & Menzies, R. (2009). Using syllable-timed speech to treat preschool children who stutter: a multiple baseline experiment. *Journal of Fluency Disorders, 34*(1), 1-10. doi:10.1016/j.jfludis.2009.01.001

Tumanova, V., Zebrowski, P. M., Throneburg, R., & Kulak Kayikci, M. E. (2011). Articulation rate and its relationship to disfluency type, duration, and temperament in preschool children who stutter. *Journal of Communication Disorders, 44*(1), 116-129. doi:10.1016/j.jcomdis.2010.09.001

Usler, E. R., & Walsh, B. (2018). The Effects of Syntactic Complexity and Sentence Length on the Speech Motor Control of School-Age Children Who Stutter. *Journal of Speech Language and Hearing Research, 61*(9), 2157-2167. doi:10.1044/2018_JSLHR-S-17-0435.

Van Borsel, J., & Tetnowski, J. A. (2007). Fluency disorders in genetic syndromes. *Journal of Fluency Disorders, 32*(4), 279-296. doi:10.1016/j.jfludis.2007.07.002

Van Borsel, J., Van Der Made, S., & Santens, P. (2003). Thalamic stuttering: A distinct clinical entity? *Brain and Language, 85*(2), 185-189. doi:10.1016/s0093-934x(03)00061-0

van de Vorst, R., & Gracco, V. L. (2017). Atypical non-verbal sensorimotor synchronization in adults who stutter may be modulated by auditory feedback. *Journal of Fluency Disorders, 53*, 14-25. doi:10.1016/j.jfludis.2017.05.004

Van Riper, C. (1971). *The nature of stuttering*. Englewood Cliffs, NJ: Prencice-Hall.

Van Riper, C. (1982). *The nature of stuttering* (2 ed.). Englewood Cliffs, NJ: Prentice-Hall.

van Riper, C. (1986). *Die Behandlung des Stotterns*. Solingen: Bundesvereinigung Stotterer-Selbsthilfe.

van Riper, C., & Emerick, L. (1984). *Speech Correction: Principles and Methods* (7 ed.). Englewood Cliffs, N.J: Prentice-Hall.

Van Zaalen-op't Hof, Y., Wijnen, F., & De Jonckere, P. H. (2009). Differential diagnostic characteristics between cluttering and stuttering - Part 1. *Journal of Fluency Disorders, 34*(3), 137-154. doi:10.1016/j.jfludis.2009.07.001

von Tiling, J. (2012). Stottern - Symptome, Ätiologie, Diagnose und Therapie. *Psychotherapeut, 6*, 537-549.

von Tiling, J., Crawcour, S. C., & Hoyer, J. (2014). *Kognitive Verhaltenstherapie des Stotterns*. Stuttgart: Kohlhammer.

Wan, C. Y., Rübner, T., Hohmann, A., & Schlaug, G. (2010). The Therapeutic Effects of Singing in Neurological Disorders. *Music Perception, 27*(4), 287-295. doi:10.1525/mp.2010.27.4.287

Ward, D. (2006). *Stuttering and Cluttering: Frameworks for Unterstanding and Treatment*. Hove: Psychology Press.

Watkins, K. E., Smith, S. M., Davis, S., & Howell, P. (2008). Structural and functional abnormalities of the motor system in developmental stuttering. *Brain, 131*(1), 50-59. doi:10.1093/brain/awm241

Watkins, R. V. (2005). Language abilities of young children who stutter. In E. Yairi & N. G. Ambrose (Eds.), *Early childhood stuttering: For clinicians by clinicians* (pp. 235-251). Austin (TX): Pro-Ed.

Watkins, R. V., Yairi, E., & Ambrose, N. G. (1999). Early childhood stuttering III: initial status of expressive language abilities. *Journal of Speech Language and Hearing Research, 42*(5), 1125-1135. doi:10.1044/jslhr.4205.1125

Webster, R. (1977). *The Precision Fluency Shaping Programm: Speech reconstruction for stutterers - Clinician`s program guide*. Roanoke, VA: Communications Development Corp.

Webster, W. G. (1990). Evidence in bimanual finger tapping of an attentional component to stuttering. *Behavioural Brain Research, 37*(2), 93-100. doi:10.1016/0166-4328(90)90084-r

Wechsler, D. (2010). *Hawik-IV - Hamburg-Wechsler-Intelligenztest für Kinder - iV* (F. Petermann & U. Petermann Eds. 3 ed.). Bern: Verlag Hans Huber.

Weikert, K. (2014). Stottern. In M. Grohnfeldt (Ed.), *Grundwissen der Sprachheilpädagogik und Sprachtherapie* (pp. 237-244). Stuttgart: Kohlhammer.

Wells, B. G., & Moore, W. H. J. (1990). EEG alpha asymmetries in stutterers and non-stutterers: effects of linguistic variables on hemispheric processing and fluency. *Neuropsychologia, 28*(12), 1295-1305. doi:10.1016/0028-3932(90)90045-p

WHO. (2001). *International Classification of Functioning, Disability and Health*. Geneva: WHO (World Health Organisation).

WHO. (2016). *International Classification of Mental and Behavioral Disorders*. Geneva: WHO (World Health Organisation).

Wieland, E. A., McAuley, J. D., Dilley, L. C., & Chang, S.-E. (2015). Evidence for a rhythm perception deficit in children who stutter. *Brain and Language, 144*, 26-34. doi:10.1016/j.bandl.2015.03.008

Wiltshire, C. E. E., Chiew, M., Chesters, J., Healy, M., & Watkins, K. E. (2021). Speech movement variability in people who stutter: A vocal tract Magnetic Resonance Imanging study. *Journal of Speech, Language, and Hearing Research, 64*(7), 2438-2452. doi:10.1044/2021_JSLHR-20-00507

Wing, A. M. (2002). Voluntary timing and brain function: an information processing approach. *Brain and Cognition, 48*(1), 7.30. doi:10.1006/brcg.2001.1301

Wing, A. M., & Kristofferson, A. B. (1973a). The timing of interresponse intervals. *Perception & Psychophysics, 13*(3), 455-460. doi:10.3758/BF03205802

Wing, A. M., & Kristofferson, A. B. (1973b). Response delays and the timing of discrete motor responses. *Perception & Psychophysics, 14*(1), 5-12. doi:10.3758/BF03198607

Wingate, M. E. (1964). Recovery from stuttering. *Journal of Speech and Hearing Disorders, 29*, 312-321.

Wingate, M. E. (1976). *Stuttering: Theory and treatment*. Boston (MA): Irvington Publishers.

Wirth, G. (2000). *Sprachstörungen, Sprechstörungen, kindliche Hörstörungen - Lehrbuch für Ärzte, Logopäden und Sprachheilpädagogen* (5 ed.). Köln: Deutscher Ärzteverlag.

Wise, R. J., Greene, J., Büchel, C., & Scott, S. K. (1999). Brain regions involved in articulation. *Lancet, 353*(9158), 1057-1061. doi:10.1016/S0140-6736(98)07491-1

Wittke-Thompson, J. K., Ambrose, N. G., Yairi, E., Roe, C., Cook, E. H., Ober, C., & Cox, N. J. (2007). Genetic studies of stuttering in a founder population. *Journal of Fluency Disorders, 32*(1), 33-50. doi:10.1016/j.jfludis.2006.12.002

Wolf, M. (2008). *Proust and the Squid: The story and science of the Reading Brain*. New York, NY, USA: Harper Perennial.

Woodrow, H. (1932). The effect of rate of sequence upon the accuracy of synchronization. *Journal of Experimental Psychology 15*(4), 357-379. doi:10.1037/h0071256

Woodruff Carr, K., White-Schwoch, T., Tierney, A., Strait, D. L., & Kraus, N. (2014). Beat synchronization predicts neural speech encoding and reading readiness in preschoolers. *Proceedings of the National Academy Science of the U.S.A., 111*(40), 14559-14564. doi:10.1073/pnas.1406219111

Xuan, Y., Meng, C., Yang, Y., Zhu, C., Wang, L., Yan, Q., . . . Yu, C. (2012). Resting-state brain activity in adult males who stutter. *PLoS ONE, 7*, e30570. doi:10.1371/journal.pone.0030570

Yairi, E. (1983). The onset of stuttering in two- and three-year old children: A preliminary report. *Journal of Speech and Hearing Disorders, 48*, 171-178. doi:10.1044/jshd.4802.171

Yairi, E., & Ambrose, N. G. (1992). A longitudinal study of stuttering in children: a preliminary report. *Journal of Speech and Hearing Research, 35*(4), 755-760.

Yairi, E., & Ambrose, N. G. (1999). Early childhood stuttering I: persistency and recovery rates. *Journal of Speech Language and Hearing Research, 42*(1097-1112).

Yairi, E., & Ambrose, N. G. (2004). *Early childhood stuttering*. Austin, TX: Pro-Ed.

Yairi, E., & Ambrose, N. G. (2005). *Early childhood stuttering: For clinicans by clinicans*. Austin, TX: Pro-Ed.

Yairi, E., & Ambrose, N. G. (2013). Epidemiology of stuttering: 21st century advances. *Journal of Fluency Disorders, 38*, 66-87.

Yan, X., & Su, X. (2009). *Linear regression analysis: Theory and computing*. Singapore, Hackensack NJ: World Scientific.

Yaruss, J. S. (2007). Application of the ICF in Fluency Disorders. *Seminars in Speech and Language, 24*(4), 312-322. doi:10.1055/s-2007-986528

Yaruss, J. S., & Quesal, R. W. (2004). Stuttering and the international classification of functioning, disability, and health (ICF): An update. *Journal of Communication Disorders, 37*(1), 35-52. doi:10.1016/S0021-9924(03)00052-2

Yaruss, J. S., & Quesal, R. W. (2006). Overall Assessment of the Speaker's Experience of Stuttering (OASES): documenting multiple outcomes in stuttering treatment. *Journal of Fluency Disorders, 31*(2), 90-115. doi:10.1016/j.jfludis.2006.02.002

Yaruss, J. S., & Quesal, R. W. (2008). *OASES: Overall Assessment of the Speaekers's Experiene of Stuttering: Manual*. Bloomington, MN: Pearson.

Zackheim, C. T., & Conture, E. G. (2003). Childhood stuttering and speech disfluencies in relation to children's mean length of utterance: A preliminary study. *Journal of Fluency Disorders, 28*(2), 115-142. doi:10.1016/s0094-730x(03)00007-x

Zatorre, R. J., Chen, J. L., & Penhune, V. B. (2007). When the brain plays music: auditory-motor interactions in music perception and production. *Nature Reviews Neuroscience, 8*(7), 547-558. doi:10.1038/nrn2152

Zelaznik, H. N., Smith, A., & Franz, E. A. (1994). Motor-performance of stutterers and nonstutterers on timing and force control tasks. *Journal of Motor Behavior, 26*(4), 340-347. doi:10.1080/00222895.1994.9941690

Zimmermann, G. N. (1980). Articulatory dynamics of „fluent“ utterances of stutterers and nonstutterers. *Journal of Speech and Hearing Research, 23*(1), 95-107. doi:10.1044/jshr.2301.95

Zimmermann, P., & Fimm, B. (2002). *Testbatterie zur Aufmerksamkeitsprüfung*. Herzogenrath: PSYTEST Verlag.

Zückner, H. (2008). *Intensiv-Modifikation Stottern: Die Modifikation. Informationen für Therapeuten* (2 ed.). Neuss: Natke Verlag.

Appendix

Appendix 1. Text excerpt for the reading task (“Eine Woche voller Samstage”, Paul Maar)

Jetzt, sagte sich Herr Taschenbier, konnte es kein Zufall mehr sein: Am Sonntag Sonne. Am Montag Herr Mon mit Mohnblumen. Am Dienstag Dienst. Am Mittwoch Mitte der Woche. Am Donnerstag Donner und am Freitag frei! Deshalb saß Herr Taschenbier am Samstag erwartungsvoll in seinem Zimmer und fragte sich, was der Tag bringen würde.

Lange hatte er noch nicht gesessen, da klopfte es laut an die Tür. Herr Taschenbier hielt vor Spannung die Luft an und sagte kein Wort. Aber es war nur Frau Rotkohl, die mit einem Eimer und einem Besen ins Zimmer kam.

»Sie können wohl nicht ›Herein‹ sagen wie jeder normale Mensch?«, fragte sie und stellte den Eimer scheppernd vor Herrn Taschenbier auf den Boden.

Erschrocken zog er die Füße unter den Stuhl zurück. Er hätte gern geantwortet:
»Ein normaler Mensch kommt auch nicht ins Zimmer, wenn niemand ›Herein‹ sagt!«

Aber Herr Taschenbier war ein netter und freundlicher Herr und hasste Streit. Außerdem hatte er ein bisschen Angst vor Frau Rotkohl, weil sie fast einen Kopf größer war als er. Und darüber hinaus war sie die Zimmerwirtin und konnte ihm jederzeit kündigen. Deswegen sagte Herr Taschenbier gar nichts.

»Sie haben wohl die Sprache verloren, Herr Taschenbier?«, fragte Frau Rotkohl weiter und begann, das Zimmer auszufegen.

»Könnten Sie nicht, bitte, mein Zimmer etwas später sauber machen?«, wagte Herr Taschenbier zaghaft zu fragen.

»Gehen Sie doch spazieren, wenn es Ihnen nicht passt!«, sagte Frau Rotkohl grob. Gleich darauf kommandierte sie: »Füße hoch!«, und fuhr mit dem Besen auf Herrn Taschenbiers Beine los. Gehorsam zog er die Füße an und stellte sie auf den Stuhl, auf dem er saß.

»Sie Schmutzfink!«, schrie Frau Rotkohl, als sie das sah. »Meinen schönen Stuhl mit Schuhen treten! Sofort gehen Sie in die Küche und holen einen Lappen!«

Herr Taschenbier eilte in die Küche. Als er wiederkam, hatte Frau Rotkohl seinen Stuhl kurzerhand auf den Tisch gestellt und wischte jetzt den Boden auf. Seufzend nahm er seinen Hut, zog seine Jacke an und ging.

»Wo wollen Sie denn hin?«, rief ihm Frau Rotkohl nach. »Spazieren gehen!«

»Das sieht Ihnen ähnlich: am hellen Tag spazieren gehen, wenn andere Leute arbeiten.«

»Sie haben doch selbst gesagt, ich solle spazieren gehen«, protestierte Herr Taschenbier.

»Das sollen Sie auch, Sie Stubenhocker«, rief sie zurück. »Sie sind schon ganz bleich, weil Sie den ganzen Tag im Zimmer hocken.«

Herr Taschenbier schlug schnell die Tür zu und machte sich auf den Weg.
Es war ein schöner Samstagmorgen, die Sonne schien, und er freute sich, dass er das Geschimpfe der Frau Rotkohl nicht mehr hören musste.

An der nächsten Straßenecke stand dicht gedrängt eine Menschengruppe.
Herr Taschenbier ging neugierig darauf zu. Die Leute betrachteten etwas. Es schien nicht sehr groß zu sein, denn alle blickten mit gesenktem Kopf nach unten. Er versuchte herauszufinden, was es da zu sehen gab. Aber er war zu klein und die Leute standen zu dicht.
»Man muss den Zoo benachrichtigen. Sicher ist es dort ausgebrochen.
»Ein gewöhnlicher Mensch hält sich so etwas nicht«, sagte eine Frau, die ganz vorn stand.
Offenbar war es irgendein Tier.
»Das scheint eine Affenart zu sein«, stellte ein Mann fest.

»Affenart? Mit dem Rüssel? Sieht eher wie eine Art Frosch aus«, rief ein anderer Mann dazwischen.
»Ein Frosch kann es unmöglich sein. Das Ding hat doch feuerrote Haare. Haben Sie schon mal einen Frosch mit Haaren gesehen? Noch dazu so groß?«

Das wurde ja immer interessanter: ein Tier, das man sowohl für einen Frosch als auch für einen Affen halten konnte!

»Sie sollten sich schämen, sich so über ein kleines Kind lustig zu machen. Sie als erwachsene Menschen, pfui!«, sagte empört eine dicke Frau und sah strafend um sich.

»Ein kleines Kind? Sie sind wohl kurzsichtig«, sagte der Mann, der das Wesen für einen Affen gehalten hatte.

Aber die dicke Frau ließ sich nicht beirren. Sie beugte sich hinunter und sagte: »Wie heißt du denn, mein Kindchen?«
Herr Taschenbier konnte immer noch nichts sehen. Aber er hörte etwas. Eine helle, durchdringende Stimme sagte laut und deutlich: »Bin kein Kindchen, bäh!«

Die umstehenden Leute rissen vor Erstaunen den Mund auf.

»Das kann ja reden!«, rief ein Mann.
»Richtig deutsch«, sagte eine Frau verwundert.

Appendix 2. Wordlists for the verbal synchronization task

Maus Hund Kind Ball Bauch
Kamm Schiff Topf Schaum Keks Korb Leim Turm Lauch Tisch Lamm Schal
Kamm Schiff Topf Schaum Keks Korb Leim Turm Lauch Tisch Lamm Schal
lieb laut toll schief kurz kühl schön lang taub kalt scharf tief
lieb laut toll schief kurz kühl schön lang taub kalt scharf tief

---

Glas Schnur Frosch Brot Stuhl
Blitz Blatt Kleid Block Staub Klang Schleim Sturm Schlauch Stich Schlamm Klo
Blitz Blatt Kleid Block Staub Klang Schleim Sturm Schlauch Stich Schlamm Klo
stark schlecht klein still schlimm klug blau stumm schlau blond klar blöd
stark schlecht klein still schlimm klug blau stumm schlau blond klar blöd

--A--

Glas Schnur Frosch Brot Stuhl
Blitz Blatt Kleid Block Staub Klang Schleim Sturm Schlauch Stich Schlamm Klo
Blitz Blatt Kleid Block Staub Klang Schleim Sturm Schlauch Stich Schlamm Klo
stark schlecht klein still schlimm klug blau stumm schlau blond klar blöd
stark schlecht klein still schlimm klug blau stumm schlau blond klar blöd

---

Maus Hund Kind Ball Bauch
Kamm Schiff Topf Schaum Keks Korb Leim Turm Lauch Tisch Lamm Schal
Kamm Schiff Topf Schaum Keks Korb Leim Turm Lauch Tisch Lamm Schal
lieb laut toll schief kurz kühl schön lang taub kalt scharf tief
lieb laut toll schief kurz kühl schön lang taub kalt scharf tief

--B--

Acknowledgements

This thesis would not have been successfully completed without the help and support of various persons involved, which is why I want to thank those persons and groups specifically:

First and foremost, I want to thank all children and adolescents who participated in this study. Without their willingness to attend all testing sessions, the study could not have been realized at all. Of course, I am also very grateful that all parents agreed to the participation. The data collection was only possible due to the help of many institutions and schools involved, which is why I also want to give special thanks to all persons that made it possible for our team to reach out for our potential participants. For the group of children and adolescents who stutter, I am especially thankful that we were allowed to attend the courses of *Stärker als Stottern* by Georg Thum and Ingeborg Mayer four times for our data collection.

The support and ideas as well as the constant exchange and communication of my two supervisors, Prof. Dr. Philip Hoole and Prof. Dr. Simone Falk, helped me to always stay focused and productive. I want to express my deepest gratitude for your indispensable input and constructive feedback during this time. This journey has been a rewarding experience of learning and exploring, including a very interesting time in Montreal – thank you, Prof. Dr. Simone Dalla Bella, for the warm welcome at BRAMS.

The whole “Timing-Team” who helped during data collection and procession made it such a pleasant experience. Especially, Mona Franke was as solid as a rock in this project – thanks to all of you for helping me adapt in the phonetic world. My girls at the office as well as Miri helped me overcome my R-aversion and were always on hand with help and advice for me. Thanks for your company.

I am beyond grateful for the support of my family and friends who helped me pass rougher times and always stayed optimistic. Special thanks to my Partner and all babysitters who helped me find the time to finish this thesis.

Finally, I want to thank the DFG for the founding of the Project and the straightforward handling of my intermittence.

Closing this chapter in my life I hope that the bonds formed during this time will outlast this experience and remain strong for new chapters ahead.