The treatment of apraxia of speech

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Chapter 2

Apraxia of Speech

2.1 | Introduction

A considerable body of research has been achieved in the last four decades on theories and concepts around Apraxia of Speech (AoS). However, there remains debate about the nature of the deficit, its neuro-anatomic basis and the symptoms of AoS. This chapter deals with these issues.

2.2 | History and definition

Liepmann (1863 – 1925) was influential in neuropsychology for his contribution on apraxia and he is usually credited with the first descriptions of apraxia (Code, 1998; Goldenberg, 2003). However, Tesak and Code (2008) refer to Bouillard (1825) who presented patients reporting symptoms that indicate what modern day linguists identify as Apraxia of Speech. Bouillard described patients with motor speech disorders but he emphasised that the tongue was not impaired. Unfortunately, further descriptions of symptoms and lesion location were relatively vague. Also, Broca (1861) and Steintahl (1881) described patients who were unable to articulate in absence of paretic speech muscles. Broca used the term *aphemia* to define an inability to coordinate the movements for the articulation of syllables and words. With this term, he distinguished between speech and language disorders but his patients only produced recurring utterances, which is insufficient to characterise specific AoS symptoms. Steinthal was a linguist who observed an aphasic patient with ideomotor apraxia in his "Synopsis of Linguistics". He concluded: "This apraxia in the narrow sense is an exaggeration of aphasia" (Steinthal, 1881). Liepmann did not cite Steinthal but in his first single case study he used the term 'Apraxie der Sprachmuskeln': *Apraxia of the speech muscles* to characterise the 'motor aphasia' of his patient (Liepmann, 1900).

Darley introduced the term 'Apraxia of Speech' in 1968. He based his first definition on Liepmann's theory and he describes AoS as an

impairment of the capacity to program movements of the articulators. Subsequently, Darley, Aronson and Brown (1975) defined AoS as a programming disorder of the articulators for the 'violational production of phonemes' (p.255). This definition entails two distinct elements of AoS: (1) positioning and (2) sequencing of articulatory movements. Incorrect positioning of articulators results in incorrect speech sound productions and the latter relates to speech sounds in an inappropriate order. An important distinctive mark in this definition is that AoS concerns a disturbed speech programming, but no phonological disorders, which are abstract phonological entities. AoS is, therefore, considered a disorder at the level of phonetic encoding.

There is a considerable amount of evidence resulting in various definitions (e.g., Wertz, LaPointe, & Rosenbek, 1984; Code, 1998; Duffy, 2005; McNeill, Robin, & Schmidt, 2009; Lowit, Miller, & Kuschmann, 2014). Most recent definitions refer to a deficit in the programming of speech movements. Accordingly, AoS patients have a preserved knowledge of the phonological word form and no deficits in motor execution. Instead, transforming abstract representations of word forms into speech motor commands is disrupted.

Current studies using online methods, such as a delayed naming task, have suggested that phonological encoding impairments may co-exist with AoS. Maas, Gutiérrez and Ballard (2014), for example, propose that activation of phonological information is delayed or protracted in AoS, according to the results of their study, which used a real-time task. The distinction between phonological and phonetic encoding disorders depend on clinical symptoms that are expected from impairments at these levels of speech production, but also on fundamental theoretical issues, such as how the processing from lexical representations to articulation proceeds (Croot, Ballard, Leyton, & Hodges, 2012; Ziegler, Aichert, & Staiger, 2012; Laganaro, 2012). Therefore, Haley, Jacks, de Riesthal, Abou-Khalil and Roth (2012) emphasise that until now, there are no

clear criteria to resolve the uncertainty arising from the overlap between phonological and phonetic disorders.

In this thesis, a definition of AoS as provided by Feiken and Jonkers (2012) is used for the theoretical background. According to them, most definitions of AoS describe a disorder of speech motor programming and speech motor planning is regarded an adaptation process of articulation. This adaptation process focuses on the dynamics of articulation and the temporal and rhythmical aspects such as tempo, stress and pause. Motor planning can be disturbed, but errors in motor planning result from deficits in motor programming (Seddoh, Robin, Sim, Hageman, Moon, & Folkins, 1996). One frequently mentioned temporal characteristic of AoS is reduction in speech rate. Effects of increased vowel and consonant durations have been observed in multisyllabic words and phrases (Kent & Rosenbek, 1983). According to Feiken and Jonkers (2012), these abnormalities in speech rate are caused by a disturbed process of speech motor programming resulting in an abnormal phoneme realisation and are not directly caused by a deficit in the process of speech motor planning. Figure 2.1 represents the underlying disorder of AoS in reference to the model of speech production including speech motor control.

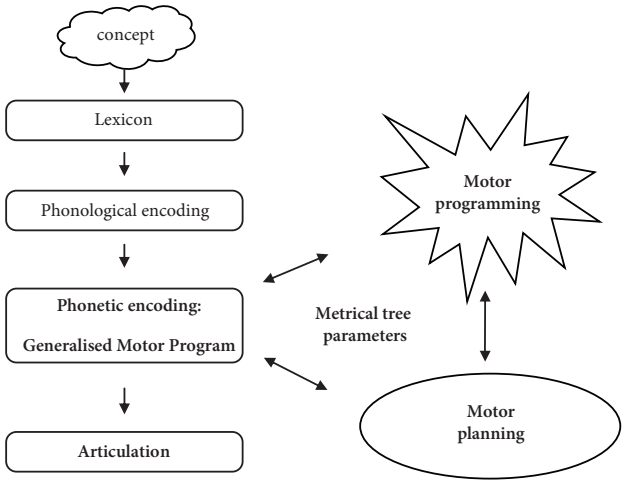

Figure 2.1 | Speech motor programming disorder of AoS

Various factors influence the error pattern in AoS. First, the influence of syllable frequency and complexity has been described (Aichert & Ziegler 2004, 2013; Staiger & Ziegler; 2008). Aichert and Ziegler (2004), for example, studied syllable-frequency effects as well as the influence of syllable structure in AoS patients on a word-repetition task. They showed an influence of syllable frequency: fewer errors were found in high-frequency syllables in both control subjects and in patients with AoS. Moreover, the results demonstrated an effect of syllable structure. Aichert and Ziegler (2004) observed a difference in the productions of monosyllabic (CCVC and CVCC) versus bi-syllabic (CVC.CV) words. Accuracy of combinations of two consonants (i.e., clusters) depends on where the clusters are located: before, after or across a syllable boundary, where the latter is not an official cluster, except if one assumes that programming takes place per phoneme. Two consonants separated by a syllable boundary were less frequently reduced to a single phoneme than clusters in the onset or the coda of a syllable, which showed that AoS patients could use syllables as a programming unit. The analysis also showed that error rates decreased from syllable-onset clusters (CCVC) to coda clusters (CVCC). Staiger and Ziegler (2008) also studied the effect of syllable frequency and syllable structure in AoS in spontaneous speech. They interviewed AoS patients with questions about everyday life topics (e.g., profession). Staiger and Ziegler (2008) observed that the same factors that influenced single word tasks, syllable frequency and syllable structure, also influenced articulatory accuracy in spontaneous speech.

Second, an influence of word stress was found in AoS (Ziegler, 2005; Aichert, Büchner, & Ziegler, 2011). This effect was language specific and related to the metrical structure of a language. AoS patients show more segmental and prosodic errors in iambic words (e.g., bi-syllabic words with a weak-strong structure, such as, 'pedal') compared to trochaic words (e.g., bi-syllabic words with a strong-weak structure, such as, 'offer') in stress-timed languages, such as Dutch, German and English.

Therefore, the regular metrical pattern may facilitate word accuracy in AoS patients (Brendel & Aichert, 2014).

2.3 | Motor theories of Apraxia of Speech

Principles of motor learning with the proposed General Motor Programs (GMPs) and parameters in the Schema Theory (Schmidt, 2003; see 1.3) can be applied to impaired speech motor systems. Schema Theory focusses on motor programming in general and it is applicable to motor speech disorders, such as AoS. Considering Schmidt's (2003) model, various aspects of motor programming may be disrupted in AoS. Actually, there are three possible levels of disruption. First, activating the GMP itself may be damaged (e.g., Clark & Robin, 1998). Alternatively, the schema that supplies the parameter settings may be impaired (Kent & Rosenbek, 1983). Finally, a combination of these two factors may underlie disorders in speech motor programming. These different levels will further be explained below.

Clark and Robin (1998) suggested a damaged GMP in AoS. They examined an oral visuomotor tracking (VMT[1]) task in AoS patients and patients with conduction aphasia. The participants learned four labio-mandibular movement sequences. The amplitude of these sequences was the same, but temporal aspects of the pattern differed. Subsequently, the amplitude determined the accuracy of the GMP and the temporal aspects were related to timing and force parameters. Clark and Robin (1989) predicted that AoS patients would show reduced GMP accuracy and, in addition, impaired parameterisation. However, none of these predictions were found to be true. Clark and Robin (1989) observed an apparent dissociation: AoS patients showed impairments in the GMP itself *or* in parameterisation, but not in both. Clark and Robin, therefore, concluded that AoS patients focus more on either the GMP or the parameters.

[1]VMT is an experimental task in which participants are presented with, in this study, an animation of a jaw-movement on a monitor and they are required to imitate this movement after the target pattern had been removed from view.

A point of criticism of this study, and other VMT studies, is that VMT tasks aim at detecting problems of learning an oral motor-task rather than performing a motor skill, such as speech (Ziegler, 2002). Therefore, the use of VMT in AoS is debated and this debate relates to the discussion about the presence of a general motor deficit in AoS, which will be described in 2.2.1.

Kent and Rosenbek (1983) suggested that the timing and force parameters might be impaired in AoS. They examined acoustic descriptions of prosodic characteristics in seven AoS patients using spectrograms of conversations. The results showed prosodic abnormalities including slow speech rate, prolongations, inter-syllabic pauses and initiation difficulties. Kent and Rosenbek (1983) suggested that these error patterns were based on disturbed spatial-temporal schemata (i.e., parameters in Schmidt's model).

Apart from the Schema Theory (Schmidt, 2003), two other approaches from the motor theory have been described in the AoS literature: (1) the two-stage model of motor programming (Klapp, 2003) and (2) the DIVA model of speech processing (Guenther, Ghosh, & Tourville, 2006). These two approaches complement the theory of Feiken and Jonkers (2012) and will be described to provide deeper insight into impairments in AoS. First, Klapp (2003) proposes a model that distinguishes two separate motor programming processes. The first one prepares the *internal* (INT) spatial and temporal structure of the movement. This information is retrieved from a motor buffer, which can be defined as a short-term memory store. INT is completed prior to initiation and is sensitive to complexity: complex units, such as syllables with an initial cluster, require longer processing time. The second process *sequences* (SEQ) motor units into their correct serial order. SEQ is sensitive to the number of units (i.e., length) but not to complexity.

Klapp (2003) examined the model's validity with reaction-time analyses and initially proposed the model on the basis of finger movements

(Klapp, 1995). Subsequently, he extended the model to speech production. Klapp (2003) suggested that words may be programmed as single units and the number of syllables determines its complexity. The hypothesis was that AoS reflects impairment of the INT stage but not of the SEQ stage. Deger and Ziegler (2002) examined this hypothesis using a reaction-time paradigm. They used non-word syllable repetition with manipulations of sequence complexity (such as, 'da-da' versus 'da-ga') and length (such as 'da-da' versus 'da-da-da') to examine the INT and SEQ process, respectively. The results showed a complexity effect but no length effect on reaction times, and, therefore, seemed to confirm the hypothesis of Klapp (2003) that AoS is a disorder in the INT stage but not in the SEQ stage. Maas, Robin, Wright, & Ballard (2008) also tested Klapp's (2003) theory with two experiments using online reaction-time measures. The first experiment was a non-speech, finger-movement task, with four different key-press responses as targets. Responses differed in terms of sequence length (i.e., one or four presses) and press duration (i.e., long or short). In this experiment, patients with AoS and non-brain damaged participants were included. The second experiment involved speech movements analogous to the finger movements with the same patients as in the first experiment. All four target-responses consisted of the repetition of the syllable [ba]. Maas et al. (2008) used a reaction-time paradigm that provided two dependent measures: (1) study time (i.e., the time to prepare a motor response; INT) and (2) reaction time (i.e., time between a "go-signal" and the execution of the response; the initiation of the movement: SEQ). The data of both experiments were also consistent with the view that AoS involves an impairment of the INT stage with an intact SEQ stage.

Second, the Directions Into Velocities of Articulators model (DIVA; Guenther et al., 2006) is an acoustically based model. The process of speech motor programming in the DIVA model starts with the activation in the so-called 'Speech Sound Map' (SSM). The SSM representation activates, in turn, a feed-forward command, which results in previously

learned motor actions for speech sounds: a speech motor program. The size of a speech motor program ranges from phonemes, to syllables and even to words and phrases (Guenther et al., 2006).

Within the DIVA model, two hypotheses are formulated to specify the speech motor programming disorder in AoS. First, the 'retrieval' hypothesis states that *access* to the motor programs is disturbed: activation or selection of the SSM representation is impaired. Second, the 'damaged programs' hypothesis states that the feed-forward commands from the SSM are disturbed, resulting in a damaged speech motor program. Mailand and Maas (2013) tested the two hypotheses within the DIVA model in a delayed picture-word interference task. In the retrieval hypothesis, more speech errors and longer reaction times are expected in trials where a distractor is presented. Therefore, Mailand and Maas (2013) manipulated the phase immediately preceding speech onset by occasionally playing a distractor word over headphones. They found that reaction times of picture naming were slower in AoS patients when using a distractor. These findings were predicted by the retrieval hypothesis but not by the damaged programs hypothesis. Therefore, Mailand and Maas (2013) provide preliminary support for the hypothesis that the retrieval of speech motor programmes is impaired in AoS patients.

2.3.1 | General motor deficit?

A highly debated issue in AoS literature is whether AoS is considered a general motor programming deficit (e.g., Ballard et al., 2000; Maas et al., 2008), which involves both speech and non-speech movements, or if impairments of these movements should be considered separate (e.g., Ziegler, 2003). The suggestion that AoS involves a fundamental impairment of praxis in the articulatory motor system that crosses both speech and non-speech tasks originates from the coincidence of AoS and orofacial apraxia. Duffy (2005) observed that 69% of the AoS patients suffer from orofacial apraxia. Also, Ballard et al. (2000) emphasised that all AoS patients suffer from orofacial apraxia. Therefore, Ballard, Robin

and Folkins (2003) argue that, an overlapping neuromotor-control system controls speech and non-speech movements. However, Wertz et al. (1984) and Duffy (1995) observed AoS patients without impairments in non-speech movements and emphasise that speech and non-speech movements are controlled by separate systems. They indicated dissociation between orofacial apraxia and AoS. Ballard et al. (2000) argued, in return, that standard clinical measures of oral and speech motor programming often lack sensitivity to detect disturbances in non-speech movements, particularly in mild AoS. For example, studies using more sophisticated methods to examine non-speech motor control (i.e., VMT) showed that motor impairment in AoS patients was not restricted to speech (e.g., Clark & Robin, 1998; Robin, Jacks, Hageman, Clark, & Woodworth, 2008; Maas et al., 2008).

Ziegler (2003) prefers a distinction between speech and non-speech movements in AoS patients. He discussed this issue within two models: first, *a task-independent* model where all motor functions are controlled by a universal sensory-motor system, irrespective of their purpose and second, *a task-dependent* model that distinguishes vegetative functions (such as breathing and swallowing), emotional expression (such as smiling and laughing) and speech. Ziegler (2003) supports the task-dependent model demonstrating that both AoS and dysarthria are dissociated from non-speech motor impairments of the oral, facial, lingual, velopharyngeal and laryngeal muscles.

Furthermore, the use of VMT in AoS has been criticised in various studies (e.g., Mchenry, Minton, Wilson, & Post, 1994; Deger, Ziegler, & Wessel, 1999; Ziegler, 2003). Mchenry et al. (1994) found no relation between intelligibility and orofacial tracking abilities and concluded that speaking is too complex to be assessed by VMT. Also, Deger et al. (1999) showed that VMT accuracy was related to scores based on limb-motor measures but not with measures of speech production. Finally, also Ziegler (2003) is not convinced by VMT studies in AoS. He argues

that VMT is not sensitive to detect core symptoms of AoS because there is no evidence for a causal relationship between visuomotor skills and AoS or dysarthria. Speech motor processes, according to Ziegler (2003), involve specific sensory-motor patterns and impairments are dissociated from impairments of other motor functions of the same musculature.

To conclude, there is no consensus whether the underlying disorder of AoS is a general motor programming impairment, including orofacial apraxia or even exceed vocal tract muscles, such as finger movements (Maas et al., 2008), or that AoS is restricted to disturbances of speech motor programming. This thesis focuses on disorders of speech movements and not on non-speech movements, and, therefore, considers at least for the purpose of the current studies these two processes as separate disorders.

2.4 | Aetiologies and localisation

A neuro-anatomic representation of AoS demonstrates the association between symptoms of AoS, brain lesions and brain structures. However, there is no one-to-one mapping between a damaged brain area and AoS symptoms (Knollman-Porter, 2008).

AoS is usually associated with a stroke to the left cerebral hemisphere (Duffy, 1995; Ogar, Willock, Baldo, Wilkins, Ludy, & Dronkers, 2006; Bonilha, Moser, Rorden, Baylis, & Fridriksson, 2006). However, AoS may also result from head trauma, tumor or other neurological diseases. A number of brain areas have been associated with AoS. Figure 2.1 shows these various regions. According to classical neuronal organisation (i.e., Brodmann areas), primary motor cortical areas of the face, mouth and larynx are located in the left frontal motor cortex (Brodmann areas 4 and 6; Yorkston, Beukelman, Strand, & Hadel, 2010). Initially, lesions in these regions were mentioned as the potential cause of AoS in several case studies (e.g., Leroux, Berger, Haglund, Pilcher, & Ojemann, 1991; Dronkers & Ogar, 2004). AoS has also been associated with Broca's area

(Brodmann areas 44 and 45). Other cortical areas have been described in association with AoS, such as the parietal lobe (e.g., Square, Roy, & Martin, 1997), and subcortical regions, such as the basal ganglia and thalamus as possible origins for AoS (Miller, 2002; Peach & Tonkovich, 2003; Wambaugh & Shuster, 2008).

Dronkers (1996) was the first to associate AoS to the left insula. However, Hillis, Work, Barker, Jacobs, Breese and Maurer (2004) found no association between AoS and lesions of the left insula, anterior insula or superior tip of the pre-central gyrus of the insula, in their MRI study of 80 acute stroke patients with and without insular damage. Instead, AoS was associated with structural damage or low blood flow in the left posterior inferior frontal gyrus. Therefore, Hillis et al. (2004) suggested that the left posterior inferior frontal gyrus is crucially involved in articulation.

Ogar et al. (2006) examined the relationship between AoS severity and the extent of the lesion. They found that all patients with AoS had lesions that included the superior pre-central gyrus of the insula whereas patients without AoS did not. However, almost every AoS patient was also diagnosed with dysarthria or aphasia. Therefore, it remains impossible to associate AoS to a single brain region.

There is an inconsistency in the neuro-anatomic findings associated with AoS; contradictory findings are influenced by various definitions of AoS. Moreover, as mentioned in Chapter 1, speech motor control consists of various processes, such as activation-competition, translation and syllabification, which are in the nonlinear dynamic fashion dependent on multiple cortical and sub-cortical circuits (Miller, 2000). Hence, it seems impossible to associate AoS to one certain location in the brain (McNeill et al., 2009). The left insula is not the location of a speech programmer, but it is an important position for the junction of motor and sensory cortices where inter hemispheric pathways and loops occur.

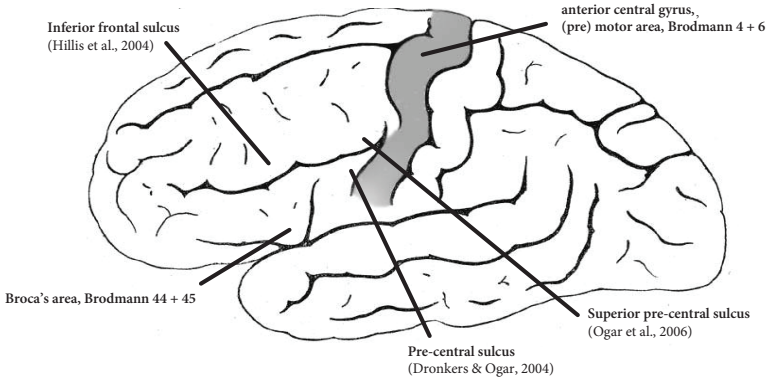

Figure 2.2 | Regions associated to AoS (adapted from www.wikipedia.org)

2.5 | Clinical presentation and diagnosis

The definition of AoS used in this thesis, as described in 2.1, relies on model-based terms, which is clinically insufficient. In addition, a description of speech characteristics completes the clinical presentation of AoS. Therefore, this paragraph discusses the many AoS symptoms.

In 1975, Darley et al. identified the following clinical features: effortful groping for articulatory movements, consonants being more affected than vowels, inconsistent error pattern, increase of errors in complex syllable structures (e.g., initial clusters), errors that approximate the target phonemes within one or two features, errors that represent perseveration, anticipation and transposition of phonemes, schwa insertions in consonant clusters and awareness of errors. Kent and Rosenbek (1983) observed disturbed timing features such as slowed speech rate with prolongation of segments, impaired coordination of voicing with movement of other articulators, and difficulty with the initiation of utterances. During the subsequent years, 33 distinctive features have been described in the literature (Sijbinga, 2009). There is partial consensus on at least some salient symptoms of AoS. Feiken and Jonkers (2012) considered the presence of eight key-symptoms of AoS in their Diagnostic Instrument for Apraxia of Speech (DIAS): (1) inconsistent realisations of phonemes, (2) more errors on consonants than on vowels,

(3) more difficulty in producing alternating syllables (e.g., "pa", "ta", "ka") than sequencing syllables (e.g., "pa", "pa", "pa"), (4) groping, (5) initiation problems, (6) segmentation of syllables, (7) segmentation of consonant clusters, and (8) effect of articulatory complexity.

In clinical practice, however, these symptoms have also been reported in patients with other neurological speech and language disorders, such as dysarthria and phonological disorders in aphasia. Therefore, no isolated symptom could serve as unambiguous inclusion criteria for diagnosing AoS (Croot, 2002). However, with a combination of symptoms it seems possible to describe AoS. Feiken and Jonkers (2012) state that the diagnosis of Dutch AoS patients, according to the DIAS, can be based on the presence of at least three of the eight mentioned symptoms. In more than 85% of the patients with speech and phonological disorders, including patients with AoS, aphasia, and dysarthria, this leads to the correct diagnosis.

Various behavioural symptoms of AoS can be classified into three categories, which will act as a distinctive mark in this thesis: impairments in accuracy, consistency and fluency (Ziegler, 2008). First, impairments in *accuracy* refer to segmental errors such as phonetic distortions. In this error type, transitions between phonemes or syllables may sound awkward and ill formed. For example, the target item [fe] may sound somewhere between a proper [fe] and [ve]. Phonemic paraphasias belong to this category as well, including (1) deletions (i.e., omission of a segment, for example "able" for "table"); (2) substitutions (i.e., replacement of a segment, for example, "cable" for "table"); (3) additions (i.e., replenishment of a segment, for example, "stable" for "table"); and (4) transpositions (i.e., conversion of two segments, for example, "battle" for "table").

Second, impairments in *consistency* refer to error variability. Johns and Darley (1970) were the first two researchers who described the variability-consistency phenomenon in AoS. Generally, error variabil-

ity serves to characterise how variable patients with AoS are in their attempts to produce speech (Staiger, Finger-Berg, Aichert, & Ziegler, 2012). Traditionally, error variability is claimed to be a key-symptom of AoS (Darley et al., 1975; Duffy, 1995; Wertz et al., 1984). However, Miller (1992) emphasised that various definitions are used on the term 'variability'. Two aspects of variability are differentiated. First, inconsistency of error *occurrence* refers to the extent to which multiple repetitions of the same target item varies in accuracy. The target item can be produced accurate in one instance and inaccurate in another. Second, inconsistency of error *types* refers to the variability of the quality of an error (e.g., phonetic distortion versus phonemic paraphasia). For example, the target item [sa] may be produced correctly in one instance, but may sound like a voiced [za] in another instance or somewhere between [sa] and [ʃa] on a third occasion. Although error variability has been a core criterion for identifying AoS since early descriptions made by Darley et al. (1975), Staiger et al. (2012) questioned the usefulness of error variability as a diagnostic criterion because there is no consensus about definition and standardised approach. Also, Haley, Jacks and Cunningham (2013) criticised the use of error variability in AoS. They found no differences in error variability in patients with AoS versus patients with aphasia with phonemic paraphasias. Accordingly, error variability should not be considered a single criterion for diagnosing AoS, but should always be connected with other symptoms of AoS. In this thesis, the DIAS is used to diagnose AoS. As described above, inconsistent realisations of phonemes is one of the eight key-symptoms of AoS in DIAS. Furthermore, consistency has been used in the evaluation instrument to measure changes in speech production during SMTA treatment. Consistency has been defined and standardisation was performed.

Finally, impairments in *fluency* refer to prosodic abnormalities, such as disturbances in the flow and melody of speech (Ziegler, 2008). These prosodic abnormalities are, next to articulatory errors, hallmarks of AoS

(Ogar, Slama, Dronkers, Amici, & Gorno-Tempini, 2005). However, it is a secondary effect of poor articulation resulting in false starts, repairs, pauses between syllables and repetitive attempts at initiating speech production. Also, prosodic impairments cause abnormal rhythm, stress and intonation patterns. These abnormalities can be assessed with the administration of screening tools, such as Motor Speech Evaluation (MSE; Wertz et al., 1984) and standardised tests for AoS, such as the Apraxia Battery for Adults (ABA-2; Dabul, 2000). For Dutch speaking AoS patients, the DIAS is used to diagnose AoS. However, dysfluent articulation in terms of the corruption of the regular rhythm and melody of speech is not defined as one of the key-symptoms in the DIAS. Therefore, a fluency measure is included in the evaluation instrument (i.e., MDT; see Chapter 8) to measure changes in prosodic features of speech production after SMTA.