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The treatment of apraxia of speech

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Chapter 9

The effectiveness of Speech-Music Therapy for Aphasia (SMTA) in five speakers with AoS and aphasia



This chapter was adapted from a peer-reviewed publication

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9.1 | Introduction

Apraxia of Speech (AoS) is a neurogenic speech motor disorder that is characterised by a wide variety of symptoms (Duffy, 2005; McNeill, Robin, & Schmidt, 2009; Lowit, Miller, & Kuschmann, 2014). The various symptoms of AoS can be classified into three categories: disorders in (a) accuracy, (b) consistency and (c) fluency (Ziegler, 2008). According to Ziegler, accuracy disorders refer to segmental impairments such as phonetic distortions and phonemic paraphasias. Disorders in consistency refer to the error variability: inaccurate productions with different qualities of the same phoneme. Finally, fluency disorders refer to prosodic impairments, such as disturbances in the flow and melody of speech, causing false starts, repairs, pauses and repetitive attempts to initiate speech production.

Various methods have been developed by speech therapists to improve verbal communication² in daily life in speakers with AoS. Wambaugh, Duffy, McNeill, Robin, & Rogers (2006) identified five general categories of AoS treatments: (1) articulatory-kinematic approaches, (2) rate-rhythm control, (3) alternative-augmentative communication, (4) intersystemic facilitation/reorganisation and (5) other. Because the treatment program under investigation (Speech-Music Therapy for Aphasia, SMTA) belongs to the rate-rhythm control strategies, we focussed on treatments in this category.

Rate-rhythm control strategies concentrate on the dynamics of articulation, such as the timing of speech production (Wambaugh & Martinez, 2000). Therapies in this category closely relate to prosodic features of articulation, and, therefore, therapies using musical elements belong to the rate-rhythm control strategies. Hurkmans, De Bruijn, Boonstra, Jonkers, Bastiaanse, Arendzen, & Reinders-Messelink (2012) reviewed

²We define verbal communication in terms of intelligibility and comprehensibility of verbal expression according to the conceptions of the Amsterdam-Nijmegen Everyday Language Test (Blomert, Kean, Koster, & Schokker, 1994).

the existing literature on the effect of music in the treatment of patients with neurological language and speech disorders. They found that Melodic Intonation Therapy (MIT; Albert, Sparks, & Helm, 1973) is one of the most studied treatment programmes. Although MIT has been regarded as a treatment for aphasia, Zumbansen, Peretz, and Hébert (2014a) recently proposed that MIT could actually be affective with respect to symptoms of AoS as well.

MIT is a hierarchically structured therapy using three main components: (a) melodic intoning, (b) rhythmic speech and (c) the use of common phrases (formulaic language). Both the melodic and the rhythmic structures are restricted to two notes (high and low) and two durations (long and short). Several studies have reported positive effects after MIT treatment, but the evidence from these studies is limited by the design: study groups have been small, and the use of variable outcome measures makes it difficult to compare these studies (Hurkmans et al., 2012; Van der Meulen, Van de Sandt-Koenderman, & Ribbers, 2012). However, Van der Meulen, Van de Sandt-Koenderman, Heijbrok-Kal, Visch-Brink, and Ribbers (2014) have recently conducted a randomised controlled clinical trial examining the efficacy of MIT in treatment of individuals with severe non-fluent aphasia during both sub-acute and chronic phases after a stroke. The results showed improved language repetition with generalisation to word retrieval and verbal communication in daily life for individuals in the subacute phase.

Although MIT uses restricted elements of music, the developers of the programme emphasised that MIT must not be seen as music therapy (MT) (Albert et al. 1973). MT is also a discipline focussing on communication, including in individuals with neurological speech and language disorders. MT is a multidisciplinary field that overlaps with several disciplines, including psychology, sociology, and neurology (Hillecke, Nickel, & Bolay, 2005). For example, neurologic MT concerns treatment of (1) cognition, such as episodic memory (Sloboda & Juslin, 2001); (2)

behaviour, such as movement disorders as in Parkinson's disease (e.g., Thaut, McIntosh, McIntosh, & Hoemberg, 2001) and (3) communication. The focus of this article is on modulation of communication that is used in neurologic MT for individuals who are not able to use language or speech, such as in individuals with AoS and aphasia. Music therapists have developed several variations of MIT, including Modified Melodic Intonation Therapy (MMIT, Baker, 2000). Apart from modifications on MIT, neurologic MT uses other programs aiming at verbal communication, such as Singen Intonation Prosodie Atmung Rhythmusübungen Improvationen (SIPARI, Jungblut & Aldridge, 2004). Within these therapies, various musical elements are used to improve speech production, such as melody, rhythm, dynamics, meter and tempo (Baker & Uhlig, 2011). However, both Hurkmans et al. (2012) and Van der Meulen et al. (2012) emphasised in their reviews that although several studies have reported improvement on speech production in individuals with AoS and aphasia with the use of musical elements, the quality of the data does not permit a firm conclusion about the benefits of MT.

Apart from above-mentioned efficacy studies, scientific attention has focussed on different therapeutic elements of MT or therapies using musical elements in neurological speech disorders, such as MIT. The production of sung and spoken utterances has been examined to assess whether the singing aids speech production for individuals with AoS and aphasia. Various experiments using different conditions, such as spoken lyrics versus lyrics sung to the original melody, and lyrics sung to a new but familiar melody, did not show that singing facilitates speech production (Hébert, Racette, Gagnon, & Peretz, 2003; Peretz, Gagnon, Hébert, & Macoir, 2004; Racette, Bard, & Peretz, 2006). This raises the question of the contribution of singing in therapies such as MIT. Recent findings suggest that singing may not be decisive but, instead, rhythm may be crucial (Stahl, Kotz, Henseler, Turner, & Geyer, 2011). Stahl, Henseler, Turner, Geyer, and Kotz (2013) found that both singing and rhythmic therapies were effective in the production of formulaic

speech in 15 individuals with global or Broca's aphasia. They, therefore, concluded that singing may not benefit speech recovery over and above rhythmic speech. Finally, Zumbansen, Peretz, and Hébert (2014b) studied the contribution of the rhythmic and pitch features of MIT. They compared melodic therapy with rhythmic therapy and spoken therapy in a cross-over design in three individuals with chronic Broca's aphasia. Their results showed, in contrast to Stahl et al. (2013), that only the melodic therapy, which consisted of both pitch and rhythm, significantly improved speech production. The above-mentioned controlled studies have demonstrated the importance of using musical elements in the efficacy of MIT, including various modifications on MIT, without leading to an understanding of which therapeutic elements, melody or rhythm, contribute to its success.

To date, all reported therapy programmes using musical elements in the treatment of AoS and aphasia are either speech therapy programmes provided by a speech therapist without the contribution of a music therapist (as in the MIT, Albert et al., 1973) or MT programmes without participation of a speech therapist (e.g., in SIPARI, Jungblut & Aldridge, 2006). However, a combination of speech and MT using all musical elements might permit a combination of the strengths of both of these therapeutic approaches: the specific knowledge of neurological speech disorders and skills of cueing strategies of the speech therapist, and the knowledge of musical parameters and specific composing skills of the music therapist. Therefore, we have developed a therapeutic approach combining elements of speech therapy and MT, which we call Speech-Music Therapy for Aphasia (SMTA, De Bruijn, Zielman, & Hurkmans, 2005). With the integration of a music therapist in SMTA, we assume that treatment effects may be maximised by adding more musical elements, and, thereby, reach variation, which seems elementary in the dynamic nature of speech motor control (Miller, 2000). Furthermore, composing melodies and selecting adequate musical elements require specific skills. A music therapist has developed these skills in contrast to

a speech therapist who might at most be musically engaged. Therefore, SMTA requires the participation of a music therapist.

In SMTA, a speech therapist and a music therapist treat the individual with AoS and aphasia *simultaneously*. The rationale for SMTA is based on similarities between language and music. A growing body of evidence from neuroimaging studies suggests that speech and music recruit shared neural systems (e.g., Brown, Martinez, & Parsons, 2006). Recent fMRI studies (Abrams, Bhatara, Ryali, Balaban, Levitin, & Menon, 2011; Rogalsky, Rong, Saberi, & Hickok, 2011) agree with the findings that music and speech processing share neural substrates but that the temporal structure in the two domains is encoded differently. Another focus of the resemblance between language and music is that both domains share hierarchical rules. Patel (2003), for example, suggested an overlap in the process of musical and linguistic syntax. Both forms of syntax relate to the connection of each incoming element X to another element Y in the evolving structure. According to Patel (2003), overlap in the processing of language and music can be conceived of as overlap in the neural areas and operations, which provide the resources for syntactic integration. Moreover, various musical elements and prosodic aspects of articulation are closely related (Stahl et al., 2011). For example, both speech and music show rhythm and intonation. In individuals with AoS, these aspects can be disturbed in speech production (Ziegler, 2008). As mentioned earlier, MIT uses two musical elements: rhythm and melody of which both underlying mechanisms are still unclear (Stahl et al., 2013; Zumbansen et al., 2014b). However, MIT structures are restricted to two notes and two durations. SMTA not only uses two notes and two durations, but maximises the spectra of melody and rhythm. Moreover, SMTA uses *all* musical elements (i.e., melody, rhythm, meter, tempo and dynamics).

SMTA consists of two interwoven lines of treatment: (a) speech therapy and (b) MT. Although the speech therapist and the music therapist

work together with the individual with AoS at the same time and in the same room, we will describe the two lines of treatment separately. The speech therapy line of treatment consists of three levels: (a) phonemes (including syllables), (b) words and (c) sentences. Trained items at the phoneme level are *always* related to the target items at the word level. For example, the syllable ‘fra’ can be trained at the phoneme level in preparation to the word “Frans”, a common Dutch name, at the word level. Furthermore, vowels can be trained in isolation. Usually, sequences of three vowels are used. These can be sequential, for example, “aa” – “aa” – “aa”, or alternating, for example, “aa” – “oo” – “ee”. Phoneme selection in the sequence is based on distinctive features of articulation. Usually, consonants are not practiced in isolation, because they may obstruct the airflow and, therefore, limit the speech fluency, but they are used in syllables. As with vowels, syllables may be trained sequentially, for example “maa” – “maa” – “maa”, or in an alternating manner, such as, “maa” – “moo” – “mee”. At this level, initial clusters can be used, for example, “smaa” – “smoo” – “smee”. The word-level exercises are designed to be functionally relevant for individuals with AoS. Names of family members, places holding meaning and other words deemed important to the individuals with AoS are trained. These personally relevant words are practiced alongside frequently occurring phrases such as “hello” (i.e., formulaic language). At the word level, common daily utterances, such as “good morning”, are trained as well. These include more than one word but are not considered a sentence. At the sentence level, selected sentences that are functionally relevant to individuals with AoS are trained. For example, “enjoy your meal” and “can you help me?”. In selecting target sentences, grammar is inferior to comprehensibility. For example, we prefer the phrase “Sit inside or outside?” instead of “Would you prefer sitting inside or outside?”.

Various cueing strategies are used by the speech therapist. These include (a) phonetic cueing (i.e., auditory presentation of the first phoneme of a syllable, word or sentence), (b) visual cueing (i.e., showing

mouth references) and (c) gestures (i.e., natural gestures supporting daily utterances, such as waving the hand while saying “goodbye”).

The musical line of treatment follows a structured progression from singing to rhythmical chanting and speaking. Each and every selected target item follows this structure. The musical interventions are designed to musically support the speech exercises, and thus, they share the same structural composition from phoneme level to word and sentence level (De Bruijn, Hurkmans, & Zielman, 2011). At the phoneme level, the music therapist uses scales or parts of scales because they are easy and known to the individual with AoS and aphasia. At the word and sentence levels, the music therapist composes *new* melodies. SMTA does not make use of familiar songs because the language output in familiar songs is usually automatically generated (Schön, Gordon, & Besson, 2005). This means that only novel melodies will be used when propositional speech is targeted. Using tempo, meter, rhythm and dynamic parameters, the music therapist is able to support the melody in order to closely follow the prosodic features of the spoken speech production. For example, the music therapist selects a 4/4 or 3/4 beat according to the stress pattern of the spoken word production.

In clinical practice, we have observed that individuals with AoS improve in their speech production after SMTA treatment. However, there is still no empirical evidence of the effect of SMTA. Therefore, the present study was a first attempt to find empirical evidence on the effect of SMTA in a small group of individuals with AoS and aphasia, as a “proof of principle”. Earlier aphasia efficacy studies (Bastiaanse, Hurkmans, & Links, 2006; Links, Hurkmans, & Bastiaanse, 2010) showed that a multiple-baseline, across-behaviours design (Fucetola, Tucker, Blank, & Corbetta, 2005) was useful to examine an experimental therapy in clinical practice. Within this design, it is possible to control for effects of spontaneous recovery, effects of generalisation without test-retest effects and follow improvement week by week. However, SMTA is a dynamic

process, because for each individual with AoS and aphasia the aims are personalised. This means that the treatment program is not structured in blocks, such as phoneme, word and sentence level, and the recovery process is expected to be gradual (i.e., not related to certain steps in the program). Therefore, we used the principle of multiple measurements before the treatment as a baseline, but excluded the various behaviours in the study design.

As the name suggests, SMTA was originally developed for non-fluent speakers with aphasia (such as Broca's aphasia and global aphasia). However, Zumbansen et al. (2014a) suggested that improved scores on language tests following MIT might be due to motor speech improvement. On the basis of our clinical judgement, we had the same idea but there was no reliable assessment tool to objectively examine changes in motor speech functioning, until Feiken and Jonkers (2012) developed a test to objectively measure AoS (Jonkers, Terband, & Maassen, 2014). Therefore, we included individuals with AoS in the current study to examine the effectiveness of SMTA treatment for AoS. The main research question was whether verbal communication in daily life improved after SMTA therapy in five individuals with AoS and aphasia after stroke. Related questions were whether accuracy, consistency, and fluency of articulation improved, whether improvement was the result of the therapy or spontaneous recovery, whether the severity of aphasia decreased and whether the improvement was stable.

9.2 | Methods

9.2.1 | Participants

Five participants were included in the study. These were the first five consecutive participants that met the inclusion criteria when the study started. The following inclusion criteria were formulated: (a) age between 18 and 75, (b) speech problems due to stroke, (c) no language or articulation disorders before stroke, (d) normal or adjusted-to-normal

hearing, (e) between 3 and 6 months poststroke, (f) diagnosis of AoS on the basis of the DIAS (Feiken & Jonkers, 2012) and (g) no previous SMTA treatment. All participants were right-handed as assessed with the “Vragenlijst voor Handvoorkeur” (Van Strien, 1992), a Dutch adaptation of the “Edinburgh Handedness Inventory” (Oldfield, 1970). None of the participants sang in a choir nor were they otherwise musically engaged.

A summary of the demographic and diagnostic data of the participants is given in Table 9.1. Diagnosis, typology of aphasia syndrome and severity of aphasia were based on the qualification of the Aachen Aphasia Test (AAT) (Graetz, De Bleser, & Willmes, 1992). Diagnosis, severity and characteristics of AoS were based on the DIAS (Feiken & Jonkers, 2012). Both tests are described in more detail in the Outcome measures section. Cognitive disorders were assessed by a neuropsychologist. Information on motor functioning of arms or legs originated from a physiotherapeutic report.

Table 9.1 Descriptive data of the participants

	participant J.V.	participant J.A.	participant J.K.	participant M.A.	participant F.P.
Gender	M	F	M	M	M
Age (in years)	68	47	72	56	49
Handedness	Right	Right	Right	Right	Right
Education	IE	IE	LE	HE	IE
Aetiology	ICVA	SAH	ICVA	HCVA	ICVA
Lesion location	MCA left	PCA left	MCA left	MCA right	MCA left
MPO	4	3	6	4	3
Diagnosis	AoS + Aphasia				
Syndrome	Broca	Broca	global	Broca	Wernicke
Severity					
AoS	mild	severe	mild	mild	severe
Aphasia	moderate	moderate	severe	moderate	severe

MPO=months post onset during pre-treatment measurements, M=male, F=female, HE = higher education, IE intermediate education, LE = lower education, ICVA = ischaemic cerebro vascular accident, HCVA=haemorrhagic cerebral vascular accident, SAH= subarachnoid haemorrhage, MCA = medial cerebral artery, PCA= posterior cerebral artery, AoS=apraxia of speech

9.2.1.1 | *Participant J.V.*

J.V. was a 68-year-old, right-handed man who had suffered a single stroke in the left middle cerebral artery. He had mild AoS that was characterised by (a) errors with consonants more than with vowels; (b) groping; (c) initiation problems and (d) segmentation of syllables. At 4 months poststroke, his aphasia was diagnosed as Broca's aphasia. His spontaneous speech was slow including phonetic distortions, phonemic paraphasias, and he was hardly intelligible. There were no other accompanying cognitive disorders than his aphasia and no motor disorders of arms or legs.

9.2.1.2 | *Participant J.A.*

J.A. was a 47-year-old, right-handed woman who had suffered a sub-arachnoid haemorrhage of the left posterior artery. She had a severe AoS with the following symptoms: (a) more difficulty in repeating alternating syllables than similar syllables on a diadochokinesis (DDK) test; (b) groping and (c) initiation problems. At 3 months poststroke, her aphasia was diagnosed as Broca's aphasia. Her spontaneous speech was hesitant and halting with many false starts, repairs and phonetic distortions. Her cognitive functioning was good except for her working memory, which was slightly reduced according to the neuropsychological report. Finally, she showed mild limb apraxia (i.e., right hand).

9.2.1.3 | *Participant J.K.*

J.K. was a 72-year-old, right-handed man who had suffered a single stroke in the left middle cerebral artery. He was diagnosed with mild AoS with the following characteristics: (a) more difficulty in repeating alternating syllables than similar syllables on a DDK test; (b) groping; (c) initiating problems and (d) segmentation of syllables. At 6 months poststroke, his aphasia was diagnosed as global aphasia. His spontaneous speech production was severely impaired. He had a hemiparesis of his right arm and no other cognitive disorders than his aphasia.

9.2.1.4 | *Participant M.A.*

M.A. was a 56-year-old, right-handed man with a haemorrhagic stroke in the middle cerebral artery in the right hemisphere. He was diagnosed with mild AoS that was characterised by (a) more articulation errors with consonants than with vowels; (b) more difficulty in repeating alternating syllables than similar syllables on a DDK test; (c) initiation problems and (d) segmentations of syllables. At 4 months poststroke, his aphasia was diagnosed as Broca's aphasia. His spontaneous speech was telegraphic: his speech rate was slow and he used predominantly content words (mainly nouns and adjectives) and he had poor morphology. The neuropsychologist reported the following cognitive disorders: (a) weak memory skills for visual stimuli; (b) decreased sustained attention; (c) slow working speed and (d) disorders in executive functioning (i.e., he was not able to rapidly switch from one task to another). His left arm and leg were paralysed.

9.2.1.5 | *Participant F.P.*

F.P. was a 49-year-old, right-handed man who had suffered from a single stroke in the left middle cerebral artery. He was diagnosed with severe AoS that was characterised by (a) articulation errors with consonants more than with vowels; (b) more difficulty in repeating alternating syllables than similar syllables on a DDK test; (c) groping and (d) segmentation of syllables. At 3 months poststroke, his aphasia was diagnosed as Wernicke's aphasia. He spoke fluently but used ill-formed sentences in his spontaneous speech, with neologisms and he was continuously attempting to repair his errors. No disorders were observed in other cognitive functioning as assessed by neuropsychological tests, nor were pareses of arms and legs present.

The participants were referred to and treated in rehabilitation centre "Revalidatie Friesland" in The Netherlands. Informed written consent was obtained from the participants prior to their inclusion in the study.

The study was approved by the Medical Ethics Committee of the University Medical Center Groningen.

9.2.2 | Outcome measures

The intelligibility score of the Amsterdam-Nijmegen Everyday Language Test (ANELT; Blomert, Koster, & Kean, 1995; maximum 50 points) was the primary outcome measure in this study since the most important goal of AoS therapy is improvement of verbal communication. The ANELT incorporates a series of daily life situations involving verbal social interaction. The verbal responses are rated on two scales: (a) comprehensibility and (b) intelligibility. The former relates to informational content and the latter relates to articulation (i.e., the degree to which the utterance can be perceived clearly).

The secondary outcome measures were: (a) The comprehensibility score of the ANELT (maximum 50 points), (b) The Token Test score on the Aachen Aphasia Test (AAT; Graetz, et al., 1992) that indicates the presence and severity of aphasia (maximum 50 points), and the repetition score of the AAT (maximum 150 points), (c) Diagnostic Instrument of Apraxia of Speech (DIAS; Feiken & Jonkers, 2012) and (d) Modified Diadochokinesis Test (MDT; Hurkmans, Jonkers, Boonstra, Stewart, & Reinders-Messelink, 2012). The DIAS and the MDT will be explained in more detail.

The DIAS is an instrument to diagnose AoS and assesses its severity (see Jonkers et al., 2014 for a more elaborate description of this test). The test consists of four subtests. In this study, three of them were used as outcome measures. The first subtest addresses articulation of phonemes. The test consists of 30 items. In this test 15 consonants and 15 vowels (including diphthongs) have to be repeated three times and the reactions are scored considering both accuracy and consistency (maximum score: 30 points). The goal of this task is to find out whether the AoS patient is able to consistently produce three identical phonemes

in a row and to establish whether more errors are made on consonants compared to vowels. The second subtest is a DDK test. This test contains 12 items; six sequencing syllables and six alternating syllables. The DDK is set up according to the level of complexity, starting with simple Consonant-Vowel (CV) structures, such as “pa”-“pa”-“pa” versus “pa”-“ta”-“ka” and ending with words with CCVCC structures, such as “stank”-“stank”-“stank” versus “tank”-“blank”-“drank”. The participant has to repeat each item as many times as possible in 8 seconds. With this test, the accuracy of repeating syllables and groping is assessed. There is no maximum score for this scale. The third test is articulation of words. This subtest contains 66 items increasing in length and articulatory complexity. The items are subdivided in 11 blocks of 6 words differing in number of syllables, number of phonemes and articulatory complexity (CVC structures, CC clusters within a syllable, CCC clusters within a syllable, CC clusters at the syllable boundary). The reactions are scored and analysed for initiation, segmentation of clusters and syllables and effects of articulatory complexity (maximum score: 264 points). In the DIAS, the diagnosis of AoS is based on the presence of eight indicative symptoms of AoS: (1) inconsistency of errors, (2) more errors with consonants than with vowels, (3) more difficulty with alternating DDK than with sequential DDK, (4) visible or audible groping, (5) initiation problems, (6) syllable segmentation, (7) segmentation of consonant clusters and (8) effects of articulatory complexity. When any three of these eight are present, a diagnosis of AoS can be secured. The authors of the DIAS compared in their validation study data of subjects with presumed AoS to subjects with dysarthria and subjects with phonological disorders in aphasia. Both the selectivity and the sensitivity of this comparison were 85%, which means that this test is good in discriminating between subjects with AoS and subjects with comparable deficits. All these symptoms are described in detail in the manual (Feiken & Jonkers, 2012) with adequate psychometric properties, such as inter-rater reliability. There are critical differences available for measuring individual improvement.

The MDT measures three symptoms of AoS: disorders in (a) consistency (i.e., error variability), (b) accuracy (i.e., segmental impairment) and (c) fluency (i.e., prosodic impairment; disturbances in the flow of speech). The MDT contains 16 pseudo-syllables. These items are divided in four blocks with the following syllable structure: (a) consonant (C) vowel (V), such as “pa”, (b) CVC, such as “paf”, (c) CVCC, such as “paks”, and (d) CCVC, such as “spag”. Each block starts with sequential diadochokinesis (e.g., “pa”-“pa”-“pa”), and then systematically alternates three distinctive features, i.e., (a) place of articulation (e.g., “pa”, “ta”, “ka”), (b) manner of articulation (e.g., “da”, “na”, “la”) and (c) vowel change (e.g., “pa”, “po”, “pu”). The individuals with AoS and aphasia were requested to repeat the examiner’s model of each string five times as accurately as possible. With regard to consistency, the maximum score is 64 points. As for accuracy, the maximum score is 240 points. Finally, as regards fluency, the maximum score is 80 points.

The control outcomes were (a) The comprehension score of spoken words and sentences of the AAT (maximum: 60 points) and (b) The score of the Dutch version of Psycholinguistic Assessment in Language Processing of Aphasia (PALPA 12) “repetition of number series”, a forward digit span task measuring short-term memory (STM) (Bastiaanse, Bosje, & Visch-Brink, 1995). The participants were requested to repeat 30 items of two, three and four number series (maximum score: 30 points). When the participants were unable to do this, they were allowed to point to the numbers on a paper.

9.2.3 | Design and procedure

We used a case series design with multiple measurements to examine treatment effects. Within this design, methodological quality was optimised. First of all, experimental control was carried out. During weekly testing, data that were related (MDT) and unrelated (PALPA 12) to the trained items were collected. Before and after treatment, an unrelated test (auditory comprehension AAT) was administered to control for

spontaneous recovery. Second, the raters participating in the analysis of the tests (both weekly testing and pre-, post-treatment, and follow-up testing) were blind to the content of the treatment, such as the trained items of phonemes, words and sentences, and all tests were in a random order when rated in a block at the end of the treatment. Third, all measures used to quantify improvement have been shown to be reliable and valid outcome measures. Finally, statistical testing was done to test whether found differences were significant.

The assessment procedure was divided into five phases. Table 9.2 provides a schematic representation of these phases.

Table 9.2 Schematic representation of the moments of administering the tests during the study

Phase	T1	Baseline	Weekly testing	T2	T3
Tests	ANELT	MDT (4 times)	MDT	ANELT	ANELT
	AAT	PALPA 12 (once)	PALPA 12	AAT	AAT
	DIAS			DIAS	DIAS

T1=pretreatment, T2=posttreatment, T3=follow-up (3 months posttreatment), AAT=Aachen Aphasia Test, DIAS=Diagnostic Instrument for Apraxia of Speech, ANELT=Amsterdam-Nijmegen Everyday Language Test, MDT=Modified Diadochokinesis Test

9.2.3.1 | *Baseline testing*

Accuracy, consistency, and fluency of articulation needed to be stable before the start of SMTA treatment to ensure that higher scores on the MDT after treatment resulted from therapy and not from test-retest effects. Therefore, the MDT was administered four times during a 2-week period prior to the first treatment session. A 10% criterion was used as a definition of stability, implying that the measures of the MDT should not exceed 10% of the consecutive scores. If there was a change of more than 10% on one of the three measures (i.e., consistency, accuracy and fluency), this had to be followed by a decrease during the following assessment.

PALPA 12 was used to measure the level of performance before the SMTA treatment as an unrelated control test. During the final baseline session, this test was administered as well.

9.2.3.2 | *Weekly testing*

During the treatment period, all participants were tested weekly with MDT and PALPA 12.

9.2.3.3 | *Pretreatment, posttreatment and follow-up testing*

Participants were tested with the ANELT, AAT and DIAS prior, 1 week and 3 months after completing the SMTA treatment. The tests were administered by a speech therapist who did not participate in the project team, or by master's students in Speech and Language Pathology (SLP) who were experienced in testing individuals with AoS and aphasia. The speech therapist providing SMTA treatment and the person administering the tests were never the same person. The assessment of the DIAS, repetition test of the AAT and ANELT were video- and audio-recorded and presented to various speech therapists and master's level students in SLP who did not know the participants. Per participant, the rater was always the same person for the different times of administration (pretreatment, and posttreatment and follow-up). The interrater agreement for DIAS and ANELT is high (Blomert et al., 1995; Feiken & Jonkers, 2012). Finally, the raters were blinded for time of administration (pretreatment or posttreatment and follow-up), and they scored the tests in random order in a block at the end of the treatment.

Posttreatment assessment was to measure generalisation of trained materials to (a) functional verbal communication (ANELT), (b) language repetition and severity of aphasia (AAT) and (c) articulation (DIAS). The follow-up assessment was to determine whether changes in verbal communication, articulation and the severity of aphasia remained stable.

9.2.4 | Treatment

9.2.4.1 | *SMTA treatment protocol*

SMTA treatment is a dynamic process. Within this process, the speech therapy line of treatment (i.e., training at the phoneme, word and sentence level) and the music therapy line of treatment (i.e., singing, rhythmical chanting and speaking, see Introduction section) take place at the same time. All musical elements are interwoven in the target items at all levels of the therapy line of treatment. This means that the treatment is not divided into blocks, such as phoneme, word and sentence level. However, there was standardisation in the treatment protocol.

The SMTA protocol comprised the following: (a) 24 treatment sessions, (b) two SMTA sessions per week and (c) 30-min treatment session. Each SMTA session started with warming up of the voice for approximately 2 minutes. Then, the speech therapy line of treatment (i.e., phoneme, word and sentence level) was followed, depending on the degree of the speech problem and target objectives. Therefore, this line of treatment was variable per participant (see Table 9.3 for the characteristics of this study). In contrast, all participants followed the same music MT line of treatment (i.e., singing, rhythmical chanting and speaking). Each target item was trained within this structure. The content of the treatment (i.e., level of the speech therapy line of treatment and the use of various musical elements) and the selection of target items were decided by the speech therapist and music therapist and were not standardised by the investigators of this study. Each target item was practised until the participant was able to produce it fluently without the therapist's help.

All participants practised the trained target items from the therapy sessions at home three times a week (i.e., on the days of the week when no SMTA therapy session was given) for half an hour. For this purpose, the target items were recorded during the therapy sessions. Recording

procedures were standardised. The participants then practised at home with CD, MP3 or another device that was suitable for them.

All participants took part in an intensive rehabilitation programme. This meant that they were receiving other interventions as well, such as physical therapy, occupational therapy, contact with a social worker, and so forth, but these never focused on speech production. Verbal communication was not trained in additional individual speech therapy. Training of auditory comprehension was excluded as well in speech therapy since this was a control task. However, reading and writing training was permitted.

9.2.5 | Statistical analysis

Kendall's tau (τ) test (with ties) was used to test for change on the MDT measures "consistency", "accuracy" and "fluency" for weekly testing during the treatment period per participant. This test establishes every week's improvement (i.e., positive correlation between two weeks resulting in a p -score of +1) or decrease (i.e., negative correlation between two weeks resulting in q -score of -1) of each measure. A tie (i.e., the same score) was scored by 0. The level of significance was set at $p < 0.05$. The Kendall test was also used to test changes on the unrelated control test (i.e., PALPA 12).

For the ANELT, AAT and DIAS critical differences for significant improvement are provided in the test manuals (Blomert et al., 1995; Feiken & Jonkers, 2012; Graetz et al., 1992) and Appendix C.6. These scores were used to evaluate whether a participant's score had significantly improved (T1 versus T2 and T2 versus T3).

9.3 | Results

All participants received 24 SMTA treatment sessions. However, the length of treatment in weeks varied per participant due to illness and holidays ranging from 12 to 20 weeks. Apart from duration, the number

of trained items for each participant varied as well due to the dynamic nature of the SMTA program. Table 9.3 provides an overview of these two characteristics.

Table 9.3 Number of trained items by the participants at all linguistic levels

	participant J.V.	participant J.A.	participant J.K.	participant M.A.	participant F.P.
Linguistic level					
Phoneme	210 (45.6)	212 (76.5)	194 (57.1)	259 (76.6)	93 (28.4)
Word	229 (49.8)	34 (12.3)	102 (30)	43 (12.4)	169 (51.5)
Sentence	21 (4.6)	31 (11.2)	44 (12.9)	37 (10)	66 (20.1)
Total	460 (100)	277 (100)	340 (100)	339 (100)	328 (100)
Length of treatment (weeks)					
	20	15	12	15	15

Number of trained items in raw scores and percentages (in brackets)

The data were analysed at the individual level. Thus, the individuals with AoS and aphasia are presented as single cases. First, intelligibility and comprehensibility of verbal communication (ANELT), articulation measures (DIAS and language repetition of the AAT), control measure (auditory comprehension AAT) and severity of the aphasia (Token Test) are described per participant directly after treatment (T2) and 3 months after treatment stopped (T3; follow-up). Second, the results of the MDT test and the control test (i.e., PALPA 12) at baseline and weekly testing during the treatment are reported (the raw scores have been transformed to percentages). Raw scores of the MDT and PALPA 12 of the weekly testing, and the raw scores of the ANELT, AAT and DIAS of T1, T2 and T3 are summarised in Appendices C.1- C.6.

The Tables show the performance patterns at T1 and T2 of the ANELT, AAT and DIAS. The figures of all participants show the results of the weekly testing³.

³The raw scores have been transformed to percentages.

9.3.1 | Participant J.V.

9.3.1.1 | ANELT, AAT and DIAS

The intelligibility of verbal communication (ANELT) changed after therapy. However, the comprehensibility of verbal communication (ANELT) showed no improvement. Articulation of phonemes (DIAS) and scores on language repetition (AAT) improved. There was no improvement on the DDK or on articulation of words (DIAS). No improvement was observed on the control task (auditory comprehension of the AAT). No improvement was seen on the Token Test (AAT) either, indicating that the SMTA treatment did not influence the severity of the aphasia. During the follow-up, no change was observed, implying that the improvement was stable, except for the intelligibility measure of the ANELT.

Table 9.4 | Performance patterns of J.V.

	ANELT		AAT			DIAS		
	intelligibility	comprehensibility	repetition	Token Test	auditory comprehension	articulation of phonemes	DDK	articulation of words
T1	+	-	+	-	-	+	-	-
T2	#	=	=	=	=	=	=	=

+ = significant improvement, # = significant decrease, - = no significant improvement, = = stable, T1 = direct after treatment, T2 = follow-up (3 months after treatment)

9.3.1.2 | Modified Diadochokinesis Test and the control test

The baseline was stable: all MDT measures met the 10% criterion. J.V. showed a high score on the fluency measure of the MDT at the first baseline testing. During further baseline assessment, there was a decrease between the first and the second assessments. The scores remained stable at assessments 3 and 4. Accuracy improved more than 10% in the final assessment but decreased at the first assessment of the weekly testing during the treatment period.

Figure 9.1 shows that during treatment all MDT measures (i.e., consistency, accuracy and fluency) gradually improved. This is most clearly visible for the consistency measure since the accuracy and fluency measure already reached levels between 70% and 80% during baseline testing. All MDT measures yielded significant improvement: consistency (Kendall $\tau = 0.58$; $p < 0.01$), accuracy (Kendall $\tau = 0.63$; $p < 0.01$) and fluency (Kendall $\tau = 0.62$; $p < 0.01$). However, the same holds for the control test: significant improvement was observed on PALPA 12 (Kendall $\tau = 0.65$; $p < 0.01$).

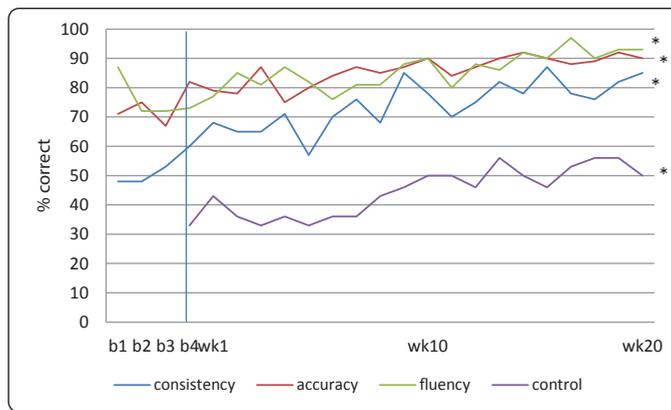


Figure 9.1 | Percentage correct production of consistency, accuracy and fluency of the MDT and repeating number series of the PALPA (control) during baseline and treatment phases of J.V., b=baseline, wk=week, $*=p < 0.05$, Kendall test with ties

9.3.2 | Participant J.A.

9.3.2.1 | ANELT, AAT and DIAS

The intelligibility of verbal communication (ANELT) changed after therapy. Also, comprehensibility of verbal communication (ANALT) and all subtests of the AAT and DIAS yielded significant improvement, except the unrelated control test for auditory comprehension of the AAT. This means that articulation improved and this improvement enhanced verbal communication. Apart from the improvement on articulation, the severity of the aphasia decreased and this is related to SMTA since there was no improvement on the control task (auditory comprehension

of the AAT). No change occurred during the follow-up testing, implying that improvement was stable.

Table 9.5 | Performance patterns of J.A.

	ANELT		AAT			DIAS		
	intelligibility	comprehensibility	repetition	Token Test	auditory comprehension	articulation of phonemes	DDK	articulation of words
T1	+	+	+	+	-	+	+	+
T2	=	=	=	=	=	=	+	=

+ = significant improvement, - = no significant improvement, = = stable, T1 = direct after treatment, T2 = follow-up (3 months after treatment)

9.3.2.2 | Modified Diadochokinesis Test and the control test

The baseline was stable: all MDT measures met the 10% criterion. Consistency improved more than 10% in the final assessment but decreased between the last baseline assessment and the first assessment of the weekly testing during treatment.

Figure 9.2 shows that all MDT measures gradually improved and reached ceiling level. Improvement was found directly after treatment for all the MDT measures: consistency (Kendall $\tau = 0.93$; $p < 0.01$), accuracy (Kendall $\tau = 0.98$; $p < 0.01$) and fluency (Kendall $\tau = 0.88$; $p < 0.01$). No improvement was seen in the control test (Kendall $\tau = 0.20$; $p > 0.05$).

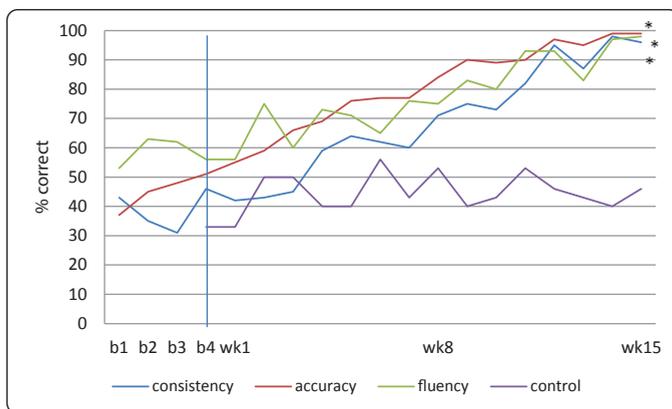


Figure 9.2 | Percentage correct production of consistency, accuracy and fluency of the MDT and repeating number series of the PALPA (control) during baseline and treatment phases of J.A., b=baseline, wk=week, * = $p < 0.05$, Kendall test with ties

9.3.3 | Participant J.K.

9.3.3.1 | ANELT, AAT and DIAS

The intelligibility of verbal communication (ANELT) changed after therapy. Also, comprehensibility of verbal communication (ANELT) and various articulation measures improved after SMTA treatment: improvement was seen on articulation of phonemes and DDK (DIAS) and repetition (AAT), except for the articulation of words (DIAS). No improvement was found on the control task (auditory comprehension of the AAT). The aphasia severity decreased: improvement was found on the Token Test (AAT). No significant changes were seen at follow-up evaluation, implying that improvement was stable.

Table 9.6 | Performance patterns of J.K.

	ANELT		AAT			DIAS		
	intelligibility	comprehensibility	repetition	Token Test	auditory comprehension	articulation of phonemes	DDK	articulation of words
T1	+	+	+	+	-	+	+	-
T2	=	=	=	=	=	=	=	=

+ = significant improvement, - = no significant improvement, = = stable, T1 = direct after treatment, T2 = follow-up (3 months after treatment)

9.3.3.2 | Modified Diadochokinesis Test and the control test

The baseline was stable: all MDT measures met the 10% criterion. Both consistency and fluency improved more than 10% between the first and the second assessments. However, this was followed by a decrease on both measures in the third assessment.

Figure 9.3 shows that the improvement was gradual for all MDT measures. Improvement was found directly after treatment for all the MDT measures: consistency (Kendall $\tau = 0.86$; $p < 0.01$), accuracy (Kendall $\tau = 0.92$; $p < 0.01$) and fluency (Kendall $\tau = 0.90$; $p < 0.01$). No significant improvement was seen on the control test (Kendall $\tau = 0.33$; $p > 0.05$).

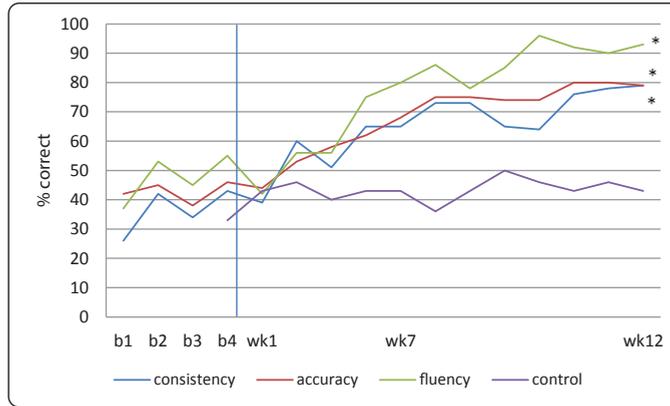


Figure 9.3 | Percentage correct production of consistency, accuracy and fluency of the MDT and repeating number series of the PALPA (control) during baseline and treatment phases of J.K., b=baseline, wk=week, $*=p<0.05$, Kendall test with ties

9.3.4 | Participant M.A.

9.3.4.1 | ANELT, AAT and DIAS

The intelligibility of verbal communication (ANELT) changed after therapy. Also, comprehensibility of verbal communication (ANELT), repetition (AAT) and DDK (DIAS) improved after SMTA. However, no improvement was found on the articulation of phonemes and words (DIAS). Improvement was seen on the Token Test, implying that the aphasia severity decreased. However, there was also improvement on the control task (auditory comprehension task of the AAT). Improvement remained stable during follow-up.

Table 9.7 | Performance patterns of M.A.

	ANELT		AAT			DIAS		
	intelligibility	comprehensibility	repetition	Token Test	auditory comprehension	articulation of phonemes	DDK	articulation of words
T1	+	+	+	+	+	-	+	-
T2	=	=	=	=	=	=	=	=

+ = significant improvement, - = no significant improvement, = = stable, T1 = direct after treatment, T2 = follow-up (3 months after treatment)

9.3.4.2 | *Modified Diadochokinesis Test and the control test*

The baseline was not stable for the fluency measure of the MDT. Fluency improved more than 10% in the final assessment and did not decrease in Week 1. However, the consistency and fluency measure were stable. Consistency improved more than 10% during the third baseline assessment but decreased during the final assessment.

Figure 9.4 shows that the improvement was gradual for all MDT measures with *consistency* and *fluency* improving the most. Significant improvement was found for all the MDT measures: consistency (Kendall $\tau = 0.65$; $p < 0.01$), accuracy (Kendall $\tau = 0.55$; $p < 0.05$) and fluency (Kendall $\tau = 0.74$; $p < 0.01$). The control test received, however, also significantly higher scores (Kendall $\tau = 0.75$; $p < 0.01$).

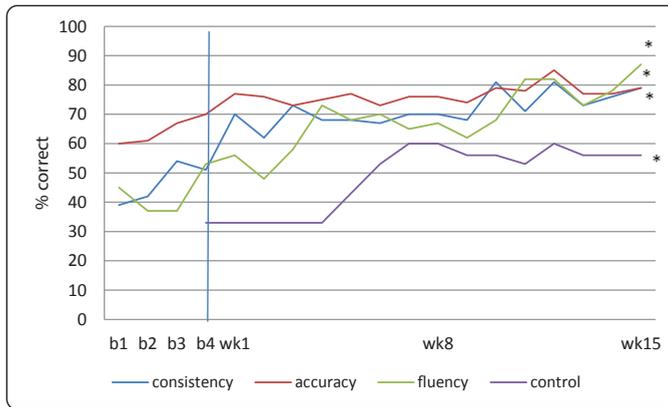


Figure 9.4 | Percentage correct production of consistency, accuracy and fluency of the MDT and repeating number series of the PALPA (control) during baseline and treatment phases of M.A., b=baseline, wk=week, *= $p < 0.05$, Kendall test with ties

9.3.5 | Participant F.P.

9.3.5.1 | *ANELT, AAT and DIAS*

The intelligibility of verbal communication (ANELT) changed after therapy. Furthermore, comprehensibility of verbal communication (ANLET) and all subtests of the AAT and DIAS improved, except the

unrelated control test for auditory comprehension (AAT). Therefore, this improvement seems to be the result of the SMTA treatment. The severity of aphasia decreased, as shown by improvement on the Token Test (AAT). The improvement was stable 3 months after therapy.

Table 9.8 | Performance patterns of F.P.

	ANELT		AAT			DIAS		
	intelligibility	comprehensibility	repetition	Token Test	auditory comprehension	articulation of phonemes	DDK	articulation of words
T1	+	+	+	+	-	+	+	+
T2	=	=	=	=	=	=	=	=

+ = significant improvement, - = no significant improvement, = = stable, T1 = direct after treatment, T2 = follow-up (3 months after treatment)

9.3.5.2 | Modified Diadochokinesis Test and the control test

The baseline was stable: all MDT measures met the 10% criterion.

Figure 9.5 shows a gradual improvement for all MDT measures, with consistency and accuracy improving the most. Improvement was significant for all the MDT measures: consistency (Kendall $\tau = 0.85$; $p < 0.01$), accuracy (Kendall $\tau = 0.96$; $p < 0.01$) and fluency (Kendall $\tau = 0.57$; $p < 0.05$). No significant improvement was found for the control test (Kendall $\tau = 0.35$; $p > 0.05$).

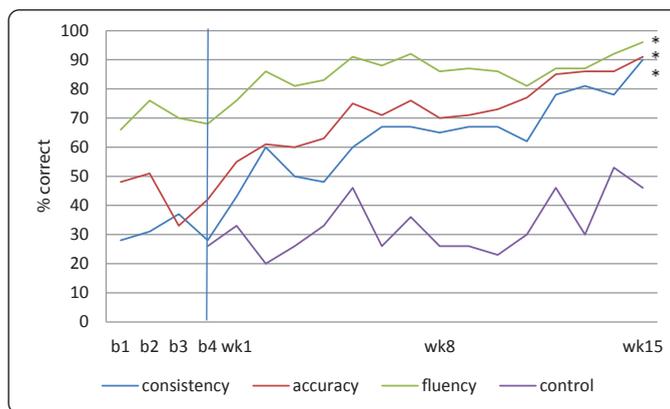


Figure 9.5 | Percentage correct production of consistency, accuracy and fluency of the MDT and repeating number series of the PALPA (control) during baseline and treatment phases of F.P., b=baseline, wk=week, * = $p < 0.05$, Kendall test with ties

9.4 | Discussion

This study was, in the first instance, a proof of principle to determine whether SMTA, without other treatment for AoS, might be useful in five individuals with AoS accompanied by aphasia. Five participants were included in a case series design with multiple measurements. The main research question was whether improvement on verbal communication in daily life was observed after SMTA therapy. Related research questions were whether accuracy, consistency and fluency of articulation improved, whether improvement was the result of the therapy or spontaneous recovery, whether the severity of the aphasia decreased and whether the improvement was stable.

Training with SMTA seemed to result in more efficient communication in daily life: intelligibility improved in all participants, and comprehensibility of verbal communication improved in four out of five participants after 24 SMTA treatment sessions. This improvement might be the result of a generalisation effect: various outcome measures related to accuracy, consistency and fluency of articulation (assessed with DIAS and MDT) showed significant change after SMTA therapy. We assume that the improvement in three out of the five participants was related to the therapy, and not to spontaneous recovery, since no improvement was found on the control tests, i.e., repetition of numbers series (PALPA 12) and auditory comprehension (AAT). These observations show that skills that were trained showed improvement (i.e., consistency, accuracy and fluency of speech production assessed with MDT) while skills that were not trained remained stable (assessed with repetition of number series assessed with PALPA 12) in these three participants.

The data showed additional findings. First, there might be generalisation to untrained related materials (DIAS) but not to untrained modalities (auditory comprehension of the AAT). This observation suggests again that higher scores on the tests were not due to spontaneous recovery. Second, SMTA not only affected articulation but also influenced

language functioning: after therapy the severity of the aphasia had decreased in four participants (as measured with the Token Test). Finally, the improvement remained stable in all participants after treatment ended (follow-up), except for the intelligibility measure of the ANELT in J.V.

Some results of this study, however, require further discussion. The first issue relates to the improvement on the PALPA 12 control test of J.V. and the improvement on both control tests of M.A. The scores of PALPA 12 of J.V. were stable until Week 8. From Week 9, he gradually improved in repeating three-number series. J.V. was ashamed that he could not accomplish this, in his own view, easy task. Therefore, he intensively tried to improve himself every week and attained 50% improvement in the following weeks. Auditory comprehension, however, did not improve, which makes us assume that the improvement on PALPA 12 is not due to spontaneous recovery, but rather to extraordinary effort made by J.V. to specifically succeed on recalling the three-number series.

However, this was not the case for M.A. His improvement seemed to be more general since all outcome measures (including both control tests) improved. Therefore, it is not sure that the improvement of M.A. can be fully related to the therapy. One can argue that his general improvement related to his “crossed aphasia”. However, De Witte, Verhoeven, Engelborghs, De Deyn, and Mariën (2008) found that the recovery of individuals with crossed aphasia is not different from the recovery of aphasia due to left hemisphere damage. Along the same lines, Benke, Bodner, and Ziegler (2011) found similar clinical patterns in a patient with right frontal lesion and AoS. Therefore, we assume that the explanation for the general improvement pattern of M.A. relates to another mechanism: improvement in sustained attention and working memory. Before the SMTA therapy, M.A. showed decreased sustained attention and slow working speed as assessed by the neuropsychologist. Although not tested after therapy, on the basis of clinical observation it was clear

that these two cognitive functions had improved and this might explain the general improvement. Whether SMTA played a role in this general cognitive improvement remains a matter for future research.

A second issue to address concerns the decrease of severity of aphasia, which was assessed with the Token Test of the AAT. The Token Test is a comprehension task: participants are requested to point to squares and circles of different colours and sizes in response to a spoken sentence. Improvement on the Token Test seems, therefore, remarkable, since auditory comprehension is not trained by SMTA. The explanation for this finding may, however, be related to *inner speech*. Inner speech is an internal monitoring process (i.e., covert speech production), and it is needed to retain auditorily presented information. The nature of inner speech is unclear. However, Vigliocco and Hartsuiker (2002) suggest that inspection of covert speech production can only be reached at end stages of the speech production process. For a successful completion of the Token Test, listeners need the process of inner speech. In part one of the Token Test, only a short sentence is given, such as “show me the red square”. However, the sentences become longer during the task. In part four, for example, participants have to respond to sentences such as “show me the small yellow square and the big green circle”. In order to respond adequately, listeners silently repeat the sentence. However, when accuracy, consistency and fluency of articulation is disrupted, as in the case of AoS, inner speech, in line with Vigliocco and Hartsuiker (2002), is also impaired. We suggest that when accuracy, consistency and fluency of articulation improve, inner speech will improve as well. Therefore, improved inner speech may explain the better scores on the Token Test.

Related to this issue is the association between STM and AoS. We selected the repetition of number series (i.e., PALPA 12) as a control test during weekly testing. This digit span task measures STM. Hickok, Rogalsky, Chen, Herskovits, Townsley and Hillis (2014) showed an

overlapping sensorimotor network (i.e., primary motor cortex, pars opercularis; pre-motor cortex and insula) between AoS and verbal STM. Therefore, one could argue that when the level of performance on STM changes, this directly influences the severity of AoS. In that case, measuring STM might not be an adequate control test. The reason for selecting a digit span task as a control task in our study was that SMTA focusses on accuracy, consistency and fluency of articulation, and not on STM. Therefore, we did not expect an effect of SMTA on STM. Results from three out of five participants of this study were consistent with this hypothesis: they improved in accuracy, consistency and fluency of articulation (assessed with MDT) and showed no improvement on STM (assessed with PALPA 12). However, two participants significantly improved in their articulation as well as their STM. For them, PALPA 12 might not have been an adequate control task, and there was no evidence of an effect of SMTA.

A third issue to address is the effect of ceiling levels. Different patterns have been observed in the success of SMTA treatment; not all participants improved significantly on all outcome measures of the different tests. A participant's ceiling level status prior to the SMTA treatment (at T1) explains lack of improvement after completing treatment. J.A., for example, scored 55 points on auditory comprehension of the AAT where 60 points is the maximum score. Significant improvement (a change of 22 points) is then not feasible. Therefore, in the end, auditory comprehension was an inadequate control measure for J.A.

A final issue we want to address relates to SMTA candidacy. Clinically, subjects of SMTA appeared to be a homogeneous group: individuals with a lesion in the left hemisphere in the medial cerebral artery and diagnosed as having AoS with aphasia. However, this study showed that SMTA candidates were various: the AoS of F.P. co-occurred with a *fluent Wernicke's* aphasia, M.A. suffered from a brain lesion in his *right* hemisphere and the lesion of J.A. was located in the *posterior* brain region.

Therefore, the results of this study indicate that SMTA might be effective to treat accuracy, consistency and fluency of articulation independent of lesion site, location and aphasia type.

The results of our study are in line with those of various studies on the effectiveness of therapies using musical elements included in the review of Hurkmans et al. (2012) and the MIT efficacy studies of Van der Meulen et al. (2014) and Zumbansen et al. (2014b). However, some crucial aspects can be differentiated. Measurable improvement was reported in the studies included in the former review (Hurkmans et al., 2012). The methodologies of these studies, however, were not adequate: (a) with regard to study design, most studies included a case study or case series without the use of any control; (b) as for blinding, most assessors were not blinded; (c) concerning comparability, use of multiple baseline assessment before treatment was sporadic; (d) with respect to outcomes, sensitive, valid and reliable outcome measures were rarely used; (e) finally, as regards significance, *p*-values were occasionally reported or calculable. In the current study, an attempt was made to improve the methodology with respect to all mentioned quality indicators. However, the results of this study do not reach the highest level of evidence due to the design (multiple baseline in multiple cases). This is an imported difference with respect to the study of Van der Meulen et al. (2014), who conducted a well-designed randomised controlled trial (RCT) and found improvement in language repetition, word retrieval and in verbal communication in individuals with subacute non-fluent aphasia. However, no outcome measures related to AoS were included; therefore, it was impossible to draw any conclusions regarding improvement on accuracy, consistency and fluency of articulation. Zumbansen et al. (2014b) did include measures related to AoS as additional, secondary outcomes. They used a diadochokinetic rate subtest of the apraxia battery for adults (ABA2; Dabul, 2000) to determine significant change on DDK scores. Zumbansen et al. (2014b) found generalisation effects in connected speech (assessed with correct information units as pri-

mary outcome) after MIT, but no significant improvement in articulation. In contrast with Zumbansen et al. (2014b), we did find significant improvement in articulation which might support the hypothesis that the integration of music therapy in AoS treatment leads to improvement in articulation and may drive changes in verbal communication.

Despite optimised methodology of the current study, we should mention two methodological limitations. The first one concerns small sample size. Inclusion of five participants prevents generalisation of the results to universal validity. However, case series with multiple measurements fit the aim of this study best. Related designs, such as the multiple baseline, across-behaviours design, are recommended in studies on effectiveness of therapies for aphasia (Bastiaanse, et al., 2006; Links, et al., 2010; Thompson, 2006; Thompson, Shapiro, Ballard, Jacobs, Schneider, & Tait, 1997) since it includes multiple baseline assessments and follows the individual's performance on trained and untrained materials week by week. It is, therefore, useful for evidence-based AoS treatment in clinical practice as well, where improvement of individuals is the most important question to be answered.

A second methodological issue relates to treatment control. The gold standard for treatment research is that the treatment under investigation is compared to a control condition (e.g., no treatment). In the SMTA study, control was included by adding multiple baseline measurements and including related and unrelated control tests. We carefully controlled for effects of spontaneous recovery. However, all five patients received only SMTA treatment.

The present study was a first attempt to find empirical evidence on the effect of SMTA in a small group of individuals with AoS and aphasia, as a “proof of principle”. The results of this study suggest that SMTA is a promising new treatment these individuals. In future research it would be interesting to study the effectiveness of SMTA in a RCT. SMTA would then be compared to another treatment, such as MIT or a treatment in

the articulatory-kinematic approach. The use of an RCT design would improve the level of evidence. This higher level of evidence is required to demonstrate the benefits of the combination of two disciplines (MT and speech therapy) compared to a therapy provided by only one professional, such as MIT, which has already shown its efficacy at the highest level of evidence. In contrast to Zumbansen et al. (2014), we do not have explicit expectations considering the role of the different musical parameters, such as melody and rhythm, but maximise the opportunities for variation by using all possibilities of various musical elements.

9.5 | Conclusion

Intelligibility of verbal communication for all participating individuals, as well as comprehensibility in four out of five participants, improved after 24 SMTA treatment sessions. All measures of MDT and repetition of AAT showed significant improvement for all participants. Four participants also improved on the test for articulation of phonemes and the diadochokinesis test of the DIAS. Furthermore, two participants improved on the articulation of words (DIAS). The improvement remained stable after treatment ended (follow-up). For three out of the five participants no improvement was found on the control tests. Two participants also showed improvement on almost all outcome measures, but also improved on the control tests. SMTA not only affected articulation but also positively influenced the severity of the aphasia in four out of five participants.

Therefore, SMTA seems an effective treatment programme for at least three of the five individuals that were treated in the current study. This treatment not only led to better articulation, but more importantly, also to improved communication in daily life.

