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The treatment of apraxia of speech

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Chapter 10
General discussion

10.1 | Introduction

The aim of this thesis was to evaluate the effect of speech-music therapy on patients with apraxia of speech. After a review of the literature on this topic (Hurkmans et al., 2012), it was concluded that efficacy studies of AoS treatment using musical elements are scarce and the methodological quality of the studies is low. Furhermore, there were no clearly defined methods to evaluate rate and rhythm control therapies for the treatment of AoS. In this research project, three studies were performed in relation to these findings. First, available clinical data of patients that were treated with SMTA were studied to examine prognostic factors influencing speech recovery. Second, a new instrument to help assess the effects of rate and rhythm therapies in clinical trials as well as daily practice was developed (Hurkmans et al., 2012). Finally, a group of patients with AoS and aphasia followed a research protocol in order to provide empirical evidence on the effect of SMTA, as a 'proof of principle' (Hurkmans et al., 2015).

The following sections discuss the main findings of these studies and elaborate on three issues. First, the relation between speech motor control and musical parameters will be discussed. The second issue addresses the evaluation of AoS treatment in rate-rhythm control strategies. Third, the effectiveness of SMTA in patients with AoS and aphasia will be described. Next, clinical implications of MIT and SMTA are presented, and, finally, an outlook for future research is given.

10.1 | The relation between speech motor control and musical parameters

Various efficacy studies on AoS treatment showed that the use of musical elements may be effective for improving speech production at the level of speech motor programming and planning (Brendel & Ziegler, 2008; Van der Meulen et al., 2014; Zumbansen et al., 2014a). A fundamental issue relates to the question how these musical elements inter-

vene in the process of speech motor control.

Brendel and Ziegler (2008), for example, developed the Metrical Pacing Therapy (MPT) in which patients are required to synchronise their articulation with a rhythmical tone sequence presented over headphones. They showed that AoS patients improved in their articulation after MPT training. One explanation for the MPT effect, as suggested by Brendel and Ziegler (2008), was that MPT provides an external metrical frame explicitly exploiting a supra-syllabic integration mechanism. However, the authors do not specify how this mechanism is related to a model of speech motor control.

Similarly to MPT, it is suggested in this thesis that SMTA provides an external *musical* frame directed to the process of speech motor control. The working mechanism of this musical frame in the process of speech motor control, as described in Chapter 1, is visualised in Figure 10.1, and will be explained below.

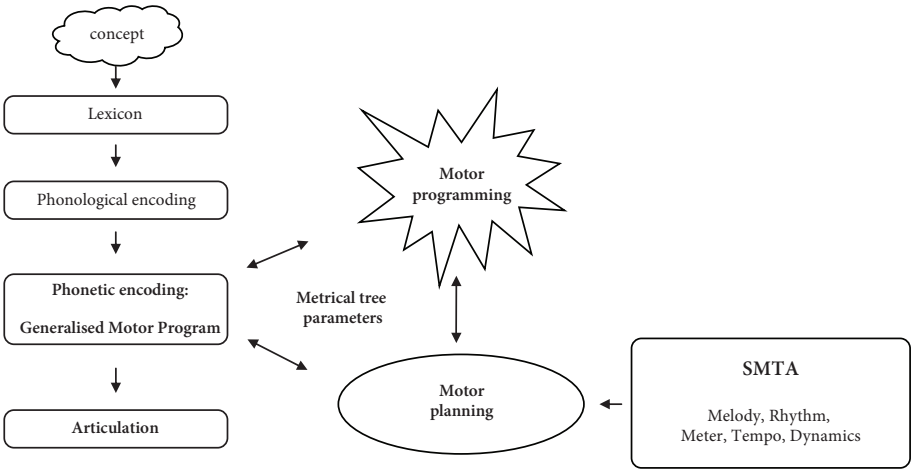

Figure 10.1 | Musical elements in relation to the model of speech motor control

SMTA uses various musical parameters to enhance a fluent speech production. Fluency characteristics refer to the flow and melody of articulation and these prosodic features are related to speech motor planning. However, Chapter 9 demonstrated that not only fluency of articu-

lation improved, but also accuracy and consistency improved after 24 SMTA treatment sessions. With this finding it can be concluded that apart from speech motor planning, SMTA also influences speech motor programming in the dynamic process of speech motor control.

The musical elements that are used in SMTA are thought to affect various aspects of speech motor control. First, melody relates to pitch. In speech, pitch is important to intonation patterns. The intonation defines, for example, whether an utterance is declarative or expresses a question. A declarative utterance is characterised by a pitch fall in prosody. In contrast, the tone of a question often rises to the top of the speaker's range (Schreuder, 2006). Timing and force parameters of Schmidt's (2003) Schema Theory determine intonation patterns in the model of speech motor control. As described in Chapter 1, these parameters are activated to determine the timing and force of a movement. For articulation, timing and force parameters are involved to determine pitch fall or rise of an utterance. Kent and Rosenbek (1983) emphasise deficient timing features in AoS that may cause disturbances in pitch fall or rise. The musical parameter 'melody' can facilitate these intonation patterns and support the linguistic prosody of an utterance.

Second, rhythm in music relates to duration: long and short. In language, rhythm is related to stress and divided into weak and strong syllables in a metrical structure. In Chapter 1, weak and strong syllables in a word structure are represented in a metrical tree. According to Ziegler (2005), rhymes and trochees are important units in the metrical tree. He found that AoS patients make more errors at unstressed syllables compared to stressed syllables. In SMTA, the music therapist composes a melody, using a rhythmic pattern that follows the stress pattern of the target item. Thus, the musical parameter, 'rhythm' supports the accuracy of all syllables, including unstressed syllables, and, thereby, prevents AoS patients from making errors.

Third, meter can be related to the musical beat. According to the descriptions of musical parameters in Chapter 5, various beats have different characters. For example, 4/4 beats are supportive whereas 3/4 beats result in a swaying motion and relaxation. These musical beats can support the gestural movements at critical positions in the word structure. As described in Chapter 1, Ziegler (2009) described transitions between neighbouring segments. He differentiates various types of vocal tract gestures, lip gestures, tongue-tip gestures and tongue-body gestures. The likelihood of accurate word production depends on the number of steps of the various gestural movements. In SMTA, the musical parameter 'meter' can strengthen these movements by selecting a beat that is adequately related to the word structure of the target item. The swaying motion of the 3/4 beat, for example, can support the transition of an initial cluster, which is difficult for AoS patients.

Fourth, the musical parameter, 'tempo' closely relates to speech rate: when the musical tempo is slow, speech rate of the lyrics in a song will also decrease. Contrarily, when the musical tempo is high, speech rate will increase. Speech motor planning can adapt articulation, such as decrease or increase of speech rate. In SMTA, the music therapist creates an opportunity for the AoS patient to articulate accurate, consistently and fluently (i.e., constant airflow) by selecting a slow musical tempo. However, a slow speech rate is artificial in verbal communication. Therefore, when accuracy, consistency and fluency of articulation improve, the musical tempo can increase as well, resulting in a more natural way of speaking.

Finally, dynamics relates to volume. Apart from linguistic prosody, such as stress and intonation patterns, emotional prosody is crucial to understand a verbal message. For example, when giving an order such as "You have to stop right now!" the emotional prosody requires the top of a speaker's volume at the end of the utterance. Processes of motor planning and the parameters of Schmidt's (2003) Schema Theory in the model of speech motor control are related to emotional prosody. The

musical parameter dynamics directly relate to these temporal aspects of articulation.

Scientific attention has focussed on the working mechanisms of these various musical elements, particularly with regard to melody and rhythm. Stahl et al. (2011) suggest that the melody may not be decisive but that rhythm is crucial. In contrast, Zumbansen et al. (2014b) showed that melodic therapy improved speech production, while rhythmic therapy did not. These contradictory findings show the lack of understanding of which therapeutic elements contribute to improvement of speech production.

For SMTA, all the various musical elements are used but it remains unknown how melody, rhythm, meter, tempo and dynamics are related to each other. The use of the five musical parameters maximises the opportunity of variation. Variation seems important in the dynamic process of speech motor control, in which accuracy, consistency and fluency of speech production depends on various aspects, such as syllable frequency and complexity (Aichert & Ziegler 2004, 2013; Staiger & Ziegler; 2008) and the metrical structure (Ziegler, 2005; Aichert, Büchner, & Ziegler, 2011), including prosodic features, such as word stress. All various musical elements can easily respond to these aspects in the process of speech motor control, but the specification of the working mechanisms can be examined in future research.

10.2 | Evaluating AoS treatment of rate and rhythm control strategies

In various studies on the effect of music in the treatment of neurological speech and language disorders, general articulation and language tests were used as outcome measures (see Chapter 5). Also, the results of the retrospective study (see Chapter 7) showed that patients with aphasia and AoS improved on language tests after SMTA in parallel with SLT. An example of a frequently used test is the AAT (Graetz et al., 1992),

which includes a repetition task for phonemes, words and sentences. However, there was no sensitive test to assess improvement in speech motor programming and, therefore, it was difficult to evaluate the effects of AoS treatment. In 2012, Feiken and Jonkers developed the DIAS, a test to diagnose AoS and assess its severity. The DIAS can also be used as an evaluation instrument to measure improvement of speech motor programming by evaluating a change in articulation of phonemes, diadochokinesis (DDK) and articulation of words after therapy. However, there was no sensitive test to help assess the effects of rate and rhythm therapies in clinical trials in a multiple baseline design, to be suitable for weekly use.

The Modified Diadochokinesis Test (MDT; Hurkmans et al., 2012) was designed with the multiple baseline design and an efficacy study of SMTA in mind. In previous studies, DDK was shown to be a sensitive variable for the assessment of speech motor programming (Ackermann et al., 1995; Ziegler, 2002). However, until now, DDK has only been used for diagnostic purposes and not for evaluation of the effects of AoS therapy. Chapter 8 describes the development of the MDT as an evaluation tool. MDT differs from classical DDK in one important aspect: omission of the variable 'speech rate'. Gadesmann and Miller (2008) reported problems regarding scoring procedures of 'rate', and the intra- and inter-rater reliabilities of DDK tests were insufficient. In contrast, the study of psychometric properties of the MDT showed that the reliability indicators were adequate. Test-retest, and intra- and inter-rater reliability were high. Validity of the MDT was adequate. Both discriminant and convergent validity showed adequate features. MDT is, therefore, an adequate instrument for efficacy studies of AoS treatment in both case studies and case-series designs.

MDT exploits the repetitive production of meaningless syllables or pseudo-syllables rather than words or sentences to evaluate the effects of speech therapy. Gadesmann and Miller (2008) emphasised the lack

of association between para-speech tasks (such as DDK-based tests) and speech tasks. The results of the study described in Chapter 8, however, showed significant correlations between MDT outcomes and those obtained with the 'classical' speech tasks, the DIAS (articulation of phonemes and words), and the ANELT, assessing functional language skills. Although Goozée et al. (2001) argued that DDK performance does not predict intelligibility, the MDT scores correlated significantly with the intelligibility measure of the ANELT.

Nevertheless, evaluating speech therapy solely by means of a para-speech task is insufficient. Therefore, speech and language tests, such as the AAT, ANELT and DIAS were used in combination with the MDT in this thesis. Thus, to study the effectiveness of AoS therapies, appropriate comprehensive and well-developed assessment strategies should be employed as pre- and post-treatment and follow-up assessments. Within this assessment strategy, the MDT can be used for the baseline measurements and the weekly assessments during the therapy period.

10.3 | Effectiveness of SMTA in patients with AoS and aphasia

This efficacy study was the first attempt to find empirical evidence on the effect of SMTA in five patients with AoS and aphasia, as a 'proof of principle' (see Chapter 9). All patients improved in their intelligibility of verbal communication in daily life. Also, comprehensibility of the functional communication improved in four patients. Apart from verbal communication, various articulation measures at the level of speech motor programming improved. In three out of the five participants it was assumed that this improvement was directly attributed to SMTA, and not to spontaneous recovery, since no improvement was found on control tests. Two participants also improved on the control tests, and, therefore, their improvement was more general, and could not only be attributed to SMTA. Additional findings revealed that the severity of aphasia decreased and improvement remained stable three months after therapy stopped.

In this thesis, the quality indicators of the ASHA level-of-evidence scheme (2001) have been used to judge the methodological quality of the efficacy study. Within this scheme nine indicators have been described (see Chapter 5) and the best methodological quality (i.e., highest score of nine points; meet all nine indicators) can only be reached within the highest level of evidence: controlled-group experiments. Using a multiple baseline design including five patients with AoS and aphasia, the SMTA efficacy study can lead to a maximum of seven points in the ASHA level-of-evidence scheme (2001). The SMTA study scored six points with the following indicators: (1) with regard to study design, control was included (i.e., no treatment control but related and unrelated measures have been collected to control for spontaneous recovery); (2) as for blinding, raters of most outcome measures were blinded; (3) concerning comparability, participants were adequately described; (4) with respect to outcomes, all used outcome measures had adequate psychometric properties (i.e., measures were valid and reliable); (5) as regards significance, statistical tests were used and *p*-values were reported, (6) finally, as for precision, effect size was reported and calculable. The following two indicators were not feasible: (1) with regard to sampling, all five patients received the same therapy (i.e., SMTA) thus randomisation was not possible; and (2) with respect to intention-to-treat, this can only be applied in randomised controlled trials. Only one feasible indicator was partly fulfilled: treatment fidelity. The research protocol prescribed two SMTA sessions per week and a total of 24 sessions. Due to illness and holidays of some of the patients, the protocol was not delivered as intended resulting in a longer duration than the intended twelve weeks.

A second methodological issue concerns sample size. The results of the SMTA efficacy study cannot be generalised to a larger population than the participants of this study. The used design (i.e., multiple baseline in case series) does not allow this. The sample size should be increased in order to generalise the results to universal validity. The gold standard for treatment research to reach such a goal is a study using an

RCT design. In the review study (see Chapter 5), none of the studies involved RCT. However, Van der Meulen et al. (2014) used an RCT design to study the efficacy of MIT in sixteen patients with non-fluent aphasia and AoS. Although the sample size is not extensive, this is the first effect-study in the rate and rhythm control strategy using the highest level of evidence with an adequate methodological quality. The results of this study showed that patients in the sub-acute phase of recovery improve in their articulation and verbal communication after MIT.

It can be concluded that both SMTA and MIT have shown to be effective in the treatment of AoS patients and patients with non-fluent aphasia. That raises the question, which program is favourited in clinical practice. The following section discusses this topic.

10.4 | SMTA and MIT in clinical practice

MIT and SMTA share similarities. Both methods (1) are hierarchically structured (focusing on singing, emphasising rhythmic speech and, finally, the intended normal speech); (2) aimed at speech motor programming and planning; (3) share patient candidacy: patients with non-fluent aphasia and AoS, including prosodic impairments in poorly articulated speech; (4) use the musical elements melody and rhythm, and are, therefore, both classified in the rate and rhythm control strategies; finally, (5) exclude the use of familiar songs.

There are differences as well. First, MIT exclusively uses functionally relevant phrases (formulaic speech). In contrast, SMTA comprises all linguistic levels (i.e., phonemes, syllables, words and sentences) using formulaic speech, for example, in utterances such as "good morning", as well as propositional speech, such as in names.

Second, the melodic and rhythmic structures in MIT are restricted to two notes (high and low) and two durations (long and short) whereas SMTA maximises the spectra of melody and rhythm. Moreover, SMTA uses all musical elements (i.e., melody, rhythm, meter, tempo and dy-

namics). Therefore, the possibilities for musical compositions are extensive with SMTA. Various musical elements can be used to compose a melody that closely relates to the word structure and features of linguistic and emotional prosody of the target words and sentences, as described in 10.1.

Finally, probably the most important difference between SMTA and MIT is the use of music *therapy* (MT) in SMTA. SMTA integrates two disciplines: speech therapy and NMT. NMT is a multidisciplinary field that overlaps with disciplines such as psychology, sociology and neurology (Hillecke et al., 2005; see Chapter 5). This means that not only the complete range of musical elements is included in SMTA but also therapeutic aspects of MT are incorporated. These aspects include: (1) with regard to action, music stimulates movement and relaxation, which can be important for speech production; (2) concerning emotion, music can affect patients, and, therefore, patients can experience feelings of joy and happiness; finally, (3) as for artistic process, during singing patients discover new possibilities that may compensate disabilities (De Bruijn et al., 2011).

However, the contribution of MT in SMTA does not automatically imply an advantage of SMTA over MIT in clinical practice. Therefore, more research is needed to broaden the knowledge about the benefits of both therapy approaches. In the next section directions for future research will be described, including a comparative study between MIT and SMTA.

10.5 | Future perspectives

The first direction for future research should be to improve the level of evidence on the efficacy of SMTA. Therefore, a study is needed in which sample size is increased and treatment type is controlled. The most adequate design for such study would be RCT with MIT as the control treatment. This seems the only appropriate procedure to reach the highest level of evidence. This higher level of evidence is required to demonstrate the benefits of the combination of two disciplines (i.e.,

MT and speech therapy) compared to a therapy provided by only one professional, such as MIT.

A second focus of future research should be directed towards the role of music therapy in SMTA. The main goal of SMTA is to improve verbal communication in daily life. Therefore, the efficacy study was designed from a linguistic perspective. However, the role of the music therapy and the use of musical elements could be studied more closely to define which components are crucial for the therapy success. As discussed earlier, Zumbansen et al. (2014b) found that melodic therapy improved speech production. However, Stahl et al. (2013) argued that it is not the singing, but the rhythm that improves speech production and singing functions as a mediator. These aspects can be studied to examine the musical parameters melody and rhythm in relation to SMTA.

Moreover, melody and rhythm have been studied most because MIT uses these two musical parameters. However, SMTA uses five musical elements. Therefore, it is possible to study all musical elements in relation to SMTA therapy and examine how they are related to each other and to the process of speech motor control.

Another suggestion for future research on SMTA would be to focus on explaining mechanisms of recovery. Until 2009, only three studies were found in the literature that included examinations of recovery mechanisms in the study's methodology to explain improvement of speech production after AoS therapy using musical elements (Belin et al., 1996; Naesser & Helm-Estabrooks, 1985; Schlaug et al., 2008). These mechanisms focused on neural correlates and, thus, used neuroimaging techniques. However, the reports are contradictory. Recent studies focus on lyric type in relation to neural correlates, which clarifies the recovery of speech. Stahl et al. (2013) proposed a two-path model of speech recovery (see Figure 10.2).

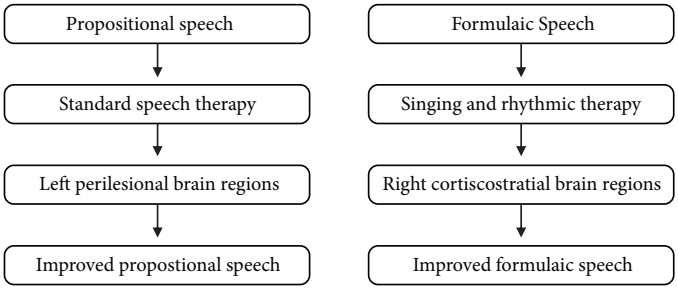

Figure 10.2 | The two-path model of speech recovery (Stahl et al., 2013)

This model could be used to understand the mechanisms of underlying treatment-induced recovery. SMTA uses both formulaic speech, in utterances such as "good morning" and propositional speech, such as "Peter, come for dinner!" In a neuroimaging study, for example using fMRI, it is possible to examine which brain areas are activated using formulaic (right corticostratial brain areas, according to Stahl et al., 2013) and propositional language (left perilesional brain areas, according to Stahl et al., 2013) during SMTA.

A final direction of future research relates to the use of transcranial stimulation. The combination of behavioural therapy with complementary brain stimulation methods is a promising direction in aphasia research: the union would further engage neural centres that are important for recovery, and facilitate neuro-plastic changes. Two techniques enhance synaptic plasticity of the brain: (1) (repetitive) Transcranial Magnetic Stimulation (rTMS) and (2) transcranial Direct Current Stimulation (tDCS). Between 2008 and 2011, ten studies testing patients with aphasia using tDCS have been performed. Vines, Norton & Schlaug (2011) studied the potential for tDCS to study the benefits of MIT. The results supported the hypothesis that combining tDSC with MIT enhances right hemisphere sensorimotor network for articulation. Hence, the advantage of using tDCS in combination with SMTA could be examined as well.

In closing, this thesis has shown that using music therapy and musical elements in combination with speech therapy is a promising direction to improve verbal communication in daily life for patients with AoS. This thesis has provided new insights into the theoretical rationale of SMTA, the evaluation of AoS treatment, and the effectiveness of SMTA. Still, there remains some potential to enhance the SMTA treatment. Suggestions for future research were given with a focus on a better understanding of the working mechanisms of SMTA and to improve the level of evidence regarding the efficacy of SMTA.