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Speech and music therapy in the treatment of CAS: An introduction and a case study

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Speech and music therapy in the treatment of CAS: An introduction and a case study

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Conflict of interest statement

The authors report no conflict(s) of interest.

Abstract

Purpose

Speech-Music Therapy for Aphasia (SMTA), a method that combines speech therapy and music therapy, is introduced as a treatment method for childhood apraxia of speech (CAS). SMTA will be evaluated in a proof-of-principle study. The first case study is presented herein.

Method

SMTA was evaluated in a study with a single-subject experimental design comparing 10 weeks of treatment with two months of no treatment. The research protocol included a pre-test, baseline phase, treatment phase, post-test, no-treatment phase and follow-up test. The participant was a five years and eight months old boy with CAS. Outcome measures were selected to reflect both intelligibility in daily communication, as well as features of CAS and speech motor planning and programming.

Results

Results on the Intelligibility in Context Scale-Dutch (ICS-Dutch) and in the analysis of a spontaneous speech sample suggest generalization of treatment effects. Improvements were found in measures that reflect complex speech motor skills, that is, the production of consonant clusters and consistency.

Conclusion

This case study showed that speech production of the participant improved after treatment with SMTA. Although intelligibility as measured with the ICS-Dutch improved over the study period, objectifying changes at the level of intelligibility in daily communication proved to be difficult. Additional measures may be necessary to gain more insight into treatment effects at this level.

Overall, the results of this first case study provide sufficient support and important leads for further evaluation of SMTA in the treatment of CAS in a proof-of-principle study.

Introduction

Childhood Apraxia of Speech (CAS) is a speech sound disorder classified as a subtype of motor speech disorder (Shriberg et al., 2010). In CAS, a core impairment at the level of speech motor planning and programming results in errors in the production of speech sounds and prosody. Inappropriate prosody, inconsistency and disrupted coarticulation are widely described as three key features of CAS (e.g., American Speech-Language-Hearing Association, 2007; Shriberg et al., 1997abc; Terband et al., 2019). Inappropriate prosody, consonant and vowel errors, and voicing and

nasality errors negatively impact intelligibility in CAS (Chenausky et al., 2022; Klopfenstein, 2009; McCabe et al., 2014), which negatively affects functional communication and social participation (Hustad, 2012).

Treatments for CAS address one or more of the three core features of CAS. In Dynamic Temporal and Tactile Cueing (DTTC; Strand, 2020; Strand et al., 2006), a method for children with more severe CAS, all three features are targeted through a focus on movements rather than phonemes, varying prosody and high numbers of repetitions. Rapid Syllable Transition treatment (ReST; Ballard et al., 2010), which is used in older children with less severe CAS, addresses all three features through the focus on sounds, beats and smoothness. Studies report positive outcomes on ratings of production accuracy for DTTC (e.g., Maas et al., 2012; Maas & Farinella, 2012; Strand et al., 2006) and on segmental accuracy and lexical stress for ReST (Ballard et al., 2012; McCabe et al., 2014). While results of these studies for different features of CAS are promising, McCabe and colleagues (2014) showed that the children improved on either lexical stress production or segmental accuracy, but they remained unable to simultaneously produce both correct stress patterns and correct segments. This is known to be a major challenge in developmental speech disorders (Howard, 2007).

Both DTTC and ReST can be described as articulatory-kinematic approaches, using interventions such as visual and tactile cues as well as feedback on the knowledge of performance. These methods also use some rate/rhythm control strategies, such as reduced speech rate and specific drill of lexical stress. Other methods, such as Melodic Intonation Therapy (MIT; Albert et al., 1973; Helfrich-Miller, 1984) and Speech-Music Therapy for Aphasia (SMTA; De Bruijn et al., 2005; Hurkmans et al., 2018), are primarily described as rate/rhythm control type approaches. The interventions in these methods are aimed at speech rate, stress, and intonation. MIT and SMTA were originally developed for adults with non-fluent aphasia and Apraxia of Speech (AoS; Hurkmans et al., 2015; Merrett et al., 2014). Both AoS and CAS are described as disorders in the planning and programming of speech movements (American Speech-Language-Hearing Association, 2007; Hurkmans, 2016) and share various characteristics, such as inconsistent errors in the realization of phonemes, syllable segregation, vowel distortions, groping and effect of articulatory complexity (Iuzzini-Seigel & Murray, 2017; Ziegler, 2008). Therefore, rate/rhythm control approaches might be effective in the treatment of CAS.

There is a limited number of studies on the use of rate/rhythm control approaches and music or musical elements in the treatment of CAS (van Tellingen et al., 2022). Four out of eight studies in the systematic review by Van Tellingen et al. (2022) evaluated the use of MIT (Helfrich-Miller, 1994; Krauss & Galloway, 1982; Lagasse, 2012; Martikainen & Korpilahti, 2011). The results of these studies vary and need to be interpreted with caution because the methodological quality of these studies was rated insufficient (van Tellingen et al., 2022).

In the present study, SMTA is evaluated in the treatment of CAS in the first case of a series in a single subject experimental design. The background and protocol for SMTA are introduced in the next section.

SMTA

SMTA is a combination of speech therapy and music therapy in which a speech language pathologist (SLP) and a music therapist (MT) provide the treatment simultaneously. It is used in clinical practice with children from three years onwards with motor speech disorders, including (suspected) CAS. This method uses musical parameters that support the prosody of speech on word, phrase, and sentence levels and facilitate the sequencing and timing of speech movements. The musical compositions are tailored to individual needs, as the music therapist composes melodies to support the functionally relevant speech targets.

There are two lines of treatment: a speech therapy line and a music therapy line, that are conducted simultaneously. Speech therapy includes three levels: (1) syllables, (2) words and (3) sentences. These levels allow for a focus on movements, rather than individual speech sounds. As an exception, vowels may be practiced in isolation at the first level. Target items on the word and sentence levels are designed both to fit the speech targets based on speech assessment, as well as to be functionally relevant (and therefore motivating) for the individual child in daily communication. For example, when a child has difficulty producing consonant clusters, and their brother's name is 'Steven', this could make this name an excellent target item. Items may be both personal, such as names of family members, and more formulaic, such as 'thank you'. Music therapy follows a structured procedure that starts with singing, followed by rhythmical chanting and speaking, which is derived from MIT. In SMTA, the final step of speaking is divided into smaller steps, including simultaneous speaking, alternating and semi-spontaneous speech. MIT uses rhythm and melody to simplify and exaggerate prosody,

limiting melody in an alternation of a limited number of pitches (Sparks, 1981). The exercises in SMTA are designed to musically support natural speech, using the musical parameters melody, rhythm, meter, tempo and dynamics. For each target item a new melody is composed to support the prosodic features of the spoken utterance. This allows for selection of targets that are specifically tailored to the communicative needs and interests, speech sound inventory, and speech motor processes of the child. During an exercise, musical parameters may be used to adjust the exercise as needed. Variations in tempo, for example, may increase or decrease the difficulty of the exercise (De Bruijn et al., 2005; Hurkmans et al., 2018). SMTA has been shown to be an effective treatment method for Dutch adults with AoS and aphasia in a proof of principle study with five speakers with aphasia and AoS (Hurkmans et al., 2015). In that study, intelligibility of verbal communication in daily life improved, as well as articulation.

SMTA is originally based on various similarities between language and music, such as shared hierarchical structures (Hurkmans, 2016; Patel, 2003; Peretz & Zatorre, 2003) and shared neural processing (e.g. Brown et al., 2006; see Hurkmans [2016] for further discussion). SMTA has been shown to improve speech production at the level of motor planning and programming (Hurkmans, 2015). To provide the rationale for SMTA in the treatment of CAS, three theoretical frameworks on music and speech and the potential working mechanisms of music in the treatment of speech will be discussed: (1) similarities between and overlap in the processing of speech and music, (2) overlap in prosodic features in music and speech, and (3) mechanisms of music with regards to motivation and mood.

One of the original fundamental ideas for SMTA is the overlap in neural processing for language and music, which can be expanded to speech. Fujii and Wan (2014) showed that an overlap in neural processing of rhythm in music and speech, combined with synchronization and entrainment to a pulse, explains how rhythm supports the recovery of speech production. Overlap between music and speech can also be found in prosody, which is characteristic for music and speech (e.g., Boutsen, 2003). Both include highly related features of sound such as melody and pitch, rhythm and duration, and dynamics and intensity (Hurkmans, 2016). Pitch, duration, and intensity are described as the features that combine to express stress in many languages (Terband et al., 2019). Through the overlap in features music can be used to support speech prosody (Hurkmans et al., 2015). The third theoretical framework concerns the positive effects of music on mood and motivation in speech-language

interventions, which have been summarized by Merrett, Peretz and Wilson (2014) as one of the possible working mechanisms of MIT. In short, music is believed to have a positive effect on mood and motivation, which may contribute to the effect of interventions that utilize musical elements. Together, these frameworks provide insight into the potential mechanisms that contribute to the effects of SMTA. SMTA encompasses all principles of motor learning that are recommended in the treatment of CAS (Maas et al., 2014; Murray et al., 2014; Strand, 2020). The use of music in SMTA allows for a high number of trials per session, which is crucial for motor learning (Maas et al., 2014; Strand, 2020), as singing (including a minimum of 20 trials per exercise) is regarded as more pleasant than realizing a high number of trials in a drill-type exercise. Usually up to five different exercises are conducted during a treatment session, alternating with small musical activities, such as singing a song or playing an instrument. Children are given autonomy within the session as they are invited to select the targets they want to practice and choose the musical activities they wish to engage in. This autonomy and the highly relevant target items can both contribute to increased motivation (Strand, 2020; Wulf et al., 2018). During an exercise, verbal feedback is kept to a minimum, to avoid disruptions of the flow of the exercise. Non-verbal knowledge of performance feedback is provided by focusing the child's attention on the provided oral example in which the speech therapist may emphasize a specific movement. Feedback in the form of knowledge of results is also provided non-verbally and focusses on accurate realizations of the target word. Placing the child's attention on accurate realizations raises their expectancies of their own ability. Before and at the end of an exercise, the functional use of the target is emphasized, to direct the attention to an external focus on results. This focus on feedback at the level of knowledge of results is recommended in the treatment of children with CAS to stimulate learning and retention (Strand, 2020) and has consistently been shown to enhance learning regardless of task, age, skill level, or (dis)ability (Wulf et al., 2018).

Treatment protocol

SMTA is provided by trained SLPs and MTs. When a child is referred for SMTA, the SLP formulates target items with the child and/ or their parents or caregivers, depending on the age of the child. These target items are at the word and/ or sentence level and are selected to be both functionally relevant for the child and relevant for the speech targets that are formulated based on speech assessment. Target items at the syllable level may be added for consonants or clusters that are still difficult for the child.

Ideally, these targets at the syllable level will subsequently be used in targets at word and/ or sentence levels.

The music therapist composes new melodies that support the natural melody, rhythm, and prosody of the target items. To this end, the music therapist uses melody, meter, rhythm, tempo, and dynamics to compose a melody that is close to the spoken prosody of the target item. This implies that musical features such as complex melodic structures, large intervals and syncopation should be avoided (for Western languages). The musical parameters can also be used to influence the difficulty of the exercise, e.g., for meter, a 6/8 beat elicits fluency more than a 4/4 beat. The composed melody consists of repetitions of the target item. The number of repetitions varies with the length of the target item, e.g., four repetitions for a sentence or eight repetitions for a word. All melodies are new and specifically composed for the target item, as famous or previously used melodies will elicit the words and sentences that go with these melodies.

During a therapy session of 30 minutes up to five target items will each be practiced in a fixed structure (see Table 1 and 2). The child is usually seated opposite of the SLP, for visual assistance, e.g., the oral example that the SLP provides. The MT is seated beside the child and SLP, creating a triangle shaped setup. The introduction of the target item by the SLP includes naming the target clearly. The introduction may be supported by an object, photograph, or picture of the target item. This is followed by a demonstration of the target item by the MT. Directly thereafter, the child and the SLP join in with the MT and sing the melody, usually two times, but more repetitions can be added if deemed necessary. The next step is rhythmic chanting. In this phase the melody fades out and the musical support is reduced to rhythmic assistance. The child and SLP can join in with the rhythmic support by tapping with the hand or foot, but this is not required. The choice to do this should be made based on what is helpful and not distracting for the child in their efforts to produce the target item. After rhythmic chanting the rhythmic support is removed and the SLP and the child simultaneously produce the target item repeatedly. Then the SLP introduces turn-taking (direct imitation) with a hand gesture, signaling when the SLP will speak and when the child is invited to speak. Finally, the SLP poses a question that will elicit the target item. In this step, any visual support used in the introductory phase may be used to repeatedly elicit the target item. During the exercises and between the phases, verbal feedback is to be kept to a minimum. Feedback may be non-verbal, with facial expression or small gestures, but interruptions of the flow of the exercise should be avoided.

Changes may be necessary during the exercise, such as a change in tempo or an extra repetition of singing. Ideally, the SLP and MT develop a cooperation so that these changes can be made during the exercise by non-verbal cues to one another, without disrupting the flow of the exercise.
Most children enjoy a break between exercises with singing a favorite song or playing an instrument for a short amount of time.

Insert table 1 and 2 around here.

When a child produces a target item correctly at semi-spontaneous (elicited) speaking during therapy, recordings of the complete exercise, can be made for practice at home. While practice at home with these recordings lacks the opportunity to provide interventions during the exercise, it does create opportunities for increasing treatment dose. Practicing at home might also contribute to the transfer of the target items to spontaneous speech in daily communication outside the treatment setting as realization of a target item in the pragmatically intended context provides a greater experience of success.

Clinical experiences with SMTA in the treatment of CAS are positive, but up to now there were no efficacy studies of SMTA in the treatment of CAS. This study represents the first single subject design study into the effectiveness of SMTA in the treatment of CAS. The main research question was whether intelligibility in daily communication improves after treatment with SMTA. Secondary research questions focused on the effect of SMTA on the production of consonants, vowels and clusters in spontaneous speech, picture naming and non-word imitation, as well as measures of speed, accuracy, consistency and fluency in a diadochokinesis task (DDK).

Case study

Method

SMTA was evaluated in a study with a single-subject experimental design comparing 10 weeks of treatment with two months of no treatment focusing on speech production (at the level of phonological encoding and speech planning and programming). The research protocol included a pre-test, baseline phase, treatment phase, post-test, no-treatment phase and follow-up test. The protocol was approved

by the research ethics committee at the University of Groningen (ref.nr. 77088008). Parents gave written informed consent prior to participation in the study. Treatment was provided at a rehabilitation center by an SLP and MT who were both trained and experienced in providing SMTA. They followed the protocol for SMTA described in the introduction. Test administrations were conducted by another SLP, who was unaware of treatment progress. All tests at all timepoints were administrated by the same SLP, at the same location, using the same equipment for test administration and recording. Recordings of the test administrations were scored by the first author, who was blinded to the order of the recordings during scoring. After scoring was completed, results were matched to their date with the key provided by the SLP who conducted the test administrations.

Participant

The participant was a 5;8 year-old Dutch-speaking boy with CAS. The diagnosis of CAS was confirmed following the protocol of Iuzzini-Seigel and Murray (2017), assessing several features of CAS in various speech tasks. The boy presented with inconsistent speech on a word and non-word repetition task and in spontaneous speech. Additional features included increasing problems with increasing complexity or length which was shown in spontaneous speech, picture naming, non-word repetition and DDK. Syllable segmentation, groping and elongation of initial consonants were observed throughout tasks. There were some consonant deletions and substitutions. In the phonological analysis of the child's spontaneous speech, the consonants /h/ and /r/ were produced accurately less than 50% of occurrences in initial position. Additionally, the consonants /s/, /d/ and /x/ (velar fricative) were produced correctly between 75% and 100% of the time in initial position. For /d/, there were several deletions in multi-syllabic words. In monosyllabic contexts, /d/ was realized accurately. In word-final position, the consonant /m/ was produced accurately in 33% of occurrences, and /l/, /k/, /n/ and /t/ were produced accurately in between 78% and 94% of occurrences. Other consonants were produced correctly in 100% of occurrences, both in initial and final position. A full overview of Dutch phonemes and this boy's phoneme acquisition is presented in appendix A. The participant had voicing difficulties, leading to whispering partial and complete utterances across speech tasks. Intelligibility was negatively influenced by suprasegmental features, such as difficulties with voicing, dysfluency, low speech rate and increasing difficulties with increasing length and/or complexity. Segmental errors impacted intelligibility to a lesser extent, with the absence of /h/ and /r/ in

his speech being striking, but also consistent. He showed awareness of his speech problems and a lack of self-confidence while speaking.

The medical history of the boy is described in detail in appendix B. His medical history included gross motor difficulties, including delayed development of walking. His fine motor skills were age appropriate and after physical therapy, his gross motor skills were age appropriate when he was 3;2 (years; months) old. He had persistent colds and tonsil issues around the age of 18 months, resulting in the clipping of his tonsils and placement of tubes in his ears around his second birthday. His hearing was within normal limits when measured at the age of 20 months and again at age 3;7.

His speech and language were assessed multiple times from the age of 2 to track progress and treatment effects. Word and sentence comprehension were within normal limits. Productive vocabulary showed delay but was within normal limits from the age of 3. Sentence production was below normal limits. Treatment was focused on increasing speech production, through targeting speech sounds and syllables. Non-verbal psychological assessment was conducted at the audiological center when the boy was 3;9, showing normal non-verbal psychological development.

The boy was placed in a specialized early education group, focusing on speech and language when he was 3;9. When he was 4;5, he went to school (which corresponds to preschool in the USA). He continued speech therapy in private practice. While there was progress in his phonological development, features of CAS became more apparent. Therefore, he was referred to the rehabilitation center for further assessment of suspected CAS and treatment at the age of 5;3.

Intervention

After the pretest, there was a two-week baseline phase. In this period five baseline measures (see below for a description of the measures) were taken. After the baseline phase treatment started, consisting of two 30-minute sessions of SMTA per week for 10 weeks, with additional homework using recordings of the targets that had been realized successfully during treatment. 10 target items on the sentence level were drawn up by the speech therapist and parents together. These items, which are presented in appendix C, were both functionally relevant to the child as well as fitting with the outcomes of his speech assessment. Items were drawn up to target both the persistent segmental errors (/r/ and /h/) and the suprasegmental features, through choosing items at the sentence level, with

multisyllabic words, and including numerous consonant clusters. Items were introduced over the treatment period as the SLP and MT saw fit.

A posttest assessment was administered after the treatment phase and a follow-up assessment was administered after a two-month no-treatment phase

Outcome measures

A schematic overview of the outcome measures and timing of administration is presented in table 3. The primary outcome measure was chosen to reflect intelligibility in daily communication, in line with the core objective of speech therapy, to support the child in communicative participation in society (Hustad, 2012). The selected measure was the Intelligibility in Context Scale – Dutch (ICS-Dutch; McLeod et al., 2012a). In this questionnaire, parents rate the intelligibility of their child in contact with various communication partners, such as family members, peers, teachers, and strangers on a five-point scale. Reliability and validity for this instrument was assessed for the original English version (McLeod et al., 2012b) and the Dutch version used in the current study (McLeod, 2020; Van Doornik et al., 2018) and found to be adequate. Both parents filled out the ICS-Dutch independent of one another.

Further outcome measures were selected to reflect speech motor abilities in various tasks, such as spontaneous speech, picture naming, non-word repetition and DDK.

A speech sample was collected and analyzed using the Phonological Analyses for Dutch (Fonologische Analyse voor het Nederlands (FAN); Beers, 1995; Beers & Masereeuw, 2022). The sample was elicited through a series of standardized questions on topics such as school, hobbies, and vacation. The first 100 unique words in the sample were transcribed and analyzed. In this task Percentage Consonants Correct in syllable initial position (PCCI) and Percentage Vowels Correct (PVC) were calculated, as well as percentage of clusters correct in syllable initial position (CCVC).

The Computer Articulation Instrument (CAI; Maassen et al., 2019) was used to assess speech in specific tasks. The CAI consists of the subtests Picture Naming, Non-word Imitation, Word- and Non-Word Repetition and DDK. For Picture Naming and Non-word Imitation PCCI, PVC and CCVC were calculated. Additionally, the occurrence of cluster reduction (Clred) was calculated. For Word- and non-wordrepetition consistency was calculated and for DDK the measure was Maximum Repetition Rate (MRR). The CAI is norm-referenced and has been shown to have sufficient to good reliability and

validity for the assessment of speech development in Dutch children ages 2-7 (van Haaften et al., 2019). The CAI norms are divided into age groups that span four months for the younger groups and six months for the two oldest groups. For this study, raw scores for the pre-test and post-test were compared with the norms for children aged 5;8-5;11 and raw scores for follow-up were compared to the norms for children aged 6;0-6;5 (van Haaften et al., 2019).

The Communication Attitude Test (CAT; Brutten, 1984; Brutten & Vanryckeghem, 2003) was used to assess the attitude of the child towards their own speech. In this norm-referenced instrument, children respond to statements about their speech with true or false. A higher score indicates a more negative attitude towards their own speech. The CAT was originally developed for children who stutter. The instrument's reliability and validity have been studied in children who stutter and were sufficient (Vanryckeghem & Brutten, 1992).

The before-mentioned tasks were assessed at pre-test, post-test and follow-up. Additionally, the Modified Diadochokinesis Test (MDT; Hurkmans et al., 2012) was used to assess speech motor planning and programming and establish a baseline and track treatment progress through weekly assessment. The MDT is a qualitative assessment of DDK. It consists of items of CV, CVC, CVCC and CCVC structure, in which the vowel, or place or manner of the consonant varies (see appendix D for the items). The child is instructed to repeat each item five times, as accurately as possible. Responses are scored for accuracy, fluency and consistency. Speech rate is not assessed. The reliability and validity of the MDT were assessed for use with adults with AoS and were sufficient for this group (Hurkmans et al., 2012). There are no records of the reliability and validity of the MDT for use with children with CAS.

A non-verbal control task was used to control for developmental progress. This was the task Figure Weights from the Wechsler Intelligence Scale for Children, Fifth Dutch edition (WISC-V-NL; Wechsler, 2018). The Figure Weights task was used in the baseline, weekly testing, and follow-up testing, alongside the MDT. Reliability and validity of the WISC-V-NL are adequate (Wechsler, 2018).

Insert table 3 around here

Analysis

Kendall's tau test was used to test for change on the MDT measures CV, CVC, CCVC, CVCC, accuracy, consistency, fluency, place, manner and vowel. The level of significance was set at $p<0.05$. Kendall's tau test was also used to analyze changes in the non-verbal control task Figure Weights. The CAI and CAT norms were used to describe changes on these measures.

Results

The participant in this study received 20 30-minute SMTA sessions, over a period of 11 weeks instead of 10, due to one week of illness during the treatment period. The number of items (both full items at the sentence level and parts of items at the word level) that was trained in each session varied from four to seven. The SLP and MT implemented repetitions of items and breaks as needed, resulting in different numbers of items practiced across sessions. In one session there were four trained items, in twelve sessions there were five, in five sessions there were six trained items and in two sessions there were seven. Each week, the items trained in the first session were repeated in the second. The first week, the more personally motivating item with the name of his stuffed animal was trained, building from practicing words to the full sentence. In the second and third week, items with the persistently difficult /r/ and /h/ were practiced at the word and sentence level. In the fourth through sixth week, items with consonant clusters were trained at the syllable, word and sentence levels. From week seven on, focus was placed on multisyllabic words, as well as consonant clusters, building up to sentence level for items containing such words. Nine out of ten of the formulated targets at sentence level were trained. The boy was able to produce the fourth item correctly upon first request during treatment. Therefore this item was not further trained. For all trained items it was necessary to practice words or syllables separately. This concerned words with consonants that were difficult for this boy, such as /h/ and /r/. Several consonant clusters were trained at the syllable level. Musical interventions were used to support speech production, such as decreased tempo for multisyllabic words and upbeats for the production of clusters. Speech therapy interventions included verbal instructions and oral examples. For example, the boy produced an interdental [n] as a substitute for /ŋ/. Verbal instruction and oral examples to keep his mouth wide open for the syllable /aŋ/ supported the production of this syllable correctly. This was subsequently integrated into the word 'belangrijk' (*important*); /ŋ/ was easier achieved in the item 'springen' (*jump*) that was trained later on. In addition to instances of feedback at the level of knowledge of performance, feedback on knowledge of results

was provided through non-verbal signs. These included signals to increase attention and effort, as well as reinforcements for adequate productions. Frequency of feedback was decreased with increasing adequate productions. Homework was provided, without fixed guidelines or expectations. Recordings of items that the boy could produce correctly during practice were sent to parents through a secure e-health application. Parents reported having had limited opportunities for practice at home. The participant received no other speech treatment at all during the second phase of this study, as this coincided with a summer break.

Results for intelligibility as measured with the ICS-Dutch, are presented in table 4. Combining the scores of both parents, the results suggested some improvement over the study period. Scores of the father and mother individually show different patterns.

Insert table 4 around here.

The analysis of a sample of spontaneous speech with the FAN suggested improvement on PCCI over the treatment period, as presented in table 5. Improvement was found for /x/ and /s/, which were realized accurate in 100% of occurrences after treatment, which was maintained at follow-up. The realization of /d/ improved after treatment and improved further over the follow-up period. For the consonant /r/ there was some improvement after treatment, which was not maintained at follow-up. Improvement on PVC and the production of initial clusters, which was obtained after treatment, was not maintained at follow-up.

Insert table 5 around here.

Tasks and measures from the CAI showed varying results. The CAI does not provide critical differences, therefore a change of Z > + 0.5 was set as the norm for clinically relevant change. Z-scores were calculated using the means and SDs published by Van Haaften et al. (2019). In Picture Naming, presented in figure 1, there was clinically relevant change for PCCI over the study period (Z + 0.51 at follow-up compared to pre-test), but no clinically relevant changes for PVC (Z + 0.42 over treatment period). The production of initial clusters in Picture Naming improved after treatment (Z + 5.46), and the gains were mostly maintained at follow-up (Z + 4.29). Results for cluster reductions in the same task, for which scores were inverted to reflect that a higher score means fewer cluster

reductions, showed clinically relevant change over the treatment period, reaching ceiling level after treatment (Z + 0.97), which was maintained at follow-up (Z + 1.16). On Non-Word Imitation, presented in figure 2, results for PCCI showed clinically relevant change after treatment (Z + 1.22), but this was not maintained at follow-up (Z + 0.16). Results for PVC showed an increase from below average to within normal limits directly after treatment (Z + 0.85), but this improvement was not maintained at follow-up (Z – 1.0). The production of initial clusters in Non-Word Imitation did not change directly after therapy, but did improve at follow-up (Z + 1.34). Results for cluster reduction in this task showed clinically relevant change, reaching ceiling level after treatment (Z + 2.8), which was maintained at follow-up (Z + 2.83). Results for consistency are presented in figure 3. Word consistency increased directly after treatment (Z + 0.80), but this improvement was not maintained at follow-up. Non-word consistency did not increase directly after treatment, but improvement was apparent at follow-up (Z + 1.71). The DDK task from the CAI, which measures maximum repetition rate (MRR; syllables per second) showed no change for the sequential items /pa/, /ta/ and /ka/. Results for the alternating items are presented in figure 4. For the items /pata/ and /taka/, results were lower directly after treatment (Z – 0.48 and Z – 2.42) and at follow-up (Z – 0.16 and Z – 0.69). For /pataka/ the scores improved after treatment (Z + 3.59) from not being able to perform this sequence at pre-test to within normal limits at follow-up (Z + 4.21).

Insert figures 1,2, 3 and 4 around here

Results for communication attitude as measured with the CAT are presented in table 6. There was no change in scores directly after treatment. At follow-up scores were lower, showing a more positive attitude towards speech.

Insert table 6 around here

Results on several measures of the MDT showed change after treatment and over the study period. For syllable structure, there was a significant gradual improvement on CV (Kendall $\tau = 0.633$, $p < .01$) and CCVC (Kendall $\tau = 0.396$, $p < .05$) structures as shown in figure 5. Results for the structures CVC (Kendall $\tau = 0.290$, $p > .05$) and CVCC (Kendall $\tau = 0.132$, $p > .05$) showed no significant change over the study period. Significant gradual improvement was found for the measures of accuracy (Kendall τ

= 0.433, $p < .05$) and consistency (Kendall $\tau = 0.447$, $p < .05$), but not for fluency (Kendall $\tau = 0.211$, $p > .05$) as shown in figure 6. Figure 7 shows significant gradual improvement for the measures place (Kendall $\tau = 0.513$, $p < .01$), manner (Kendall $\tau = 0656$, $p < .01$) and vowel (Kendall $\tau = 0.356$, $p < .05$). For the scores on the non-verbal control task Figure Weights, which is also shown in figure 7, there was no significant improvement over the study period (Kendall $\tau = -0.210$, $p > .05$).

Insert figure 5, 6 and 7 around here

Discussion

In this article we introduced Speech-Music Therapy for Aphasia (SMTA) as a new method in the treatment of childhood apraxia of speech (CAS). SMTA combines speech therapy and music therapy and is designed to support speech production at the level of motor planning and programming. Its potential for the treatment of CAS is supported by evidence of its effectiveness in adults with Apraxia of Speech (AoS; Hurkmans et al., 2015) as well as theoretical frameworks of the neural processing of rhythm in speech and music (Fujii & Wan, 2014), similarities between speech and music at the level of prosody (Hurkmans, 2016; Terband et al., 2019), and the positive effects of music on mood and motivation (Merrett et al., 2014). As a first investigation of this potential, we evaluated the effectiveness of SMTA in the treatment of CAS in a multiple baseline single subject design. First and foremost, this study showed that SMTA can be administered to five- to six-year-old children with CAS. Furthermore, the treatment yielded positive outcomes in speech production and intelligibility for the participant in this case study.

The study was designed to evaluate SMTA on a range of outcome measures, including intelligibility in daily communication and communication attitude, as well as a variety of measures of speech motor planning and programming in specific speech-motor tasks. Tasks included picture naming, non-word imitation, word- and non-word repetition, and DDK, with outcome measures such as percentage of initial consonants correct, production of clusters, consistency and fluency. Overall results showed progress on measures of speech motor planning and programming occurring directly after treatment. Improvement on intelligibility and communication attitude was mostly obtained over the entire study period and became apparent at follow-up.

As the goal of treatment in clinical practice lies in optimizing communication in daily life, we chose intelligibility in daily communication as the main outcome measure. In the scores of both parents on the ICS-Dutch combined, improvement was found at follow-up, but no effect was found directly after treatment. Examining the individual data of the two parents revealed that the father scored lower intelligibility directly after treatment. A possible explanation for this might be an increased awareness of his son's speech problems through the pre-test and treatment phases. Participation in the study and the parental questionnaires filled out at the beginning of the study might have drawn attention and raised awareness of the speech problems, leading to lower scores directly after treatment.

A similar trend was found in the scores of the CAT, with improvement in the boy's attitudes towards his own speech apparent at follow-up, but not directly after treatment. This was observed in both the results on the CAT and clinical observations by the SLP who conducted the test administrations as well as by the first author who judged and scored the video-recordings of spontaneous speech and test administrations. Clinical observations included changes in posture, communicative initiative and speaking more freely. Two underlying mechanisms may have contributed to the pattern of improvement at follow-up rather than directly after treatment. First, during treatment, focus is put on speech and the difficulties with speech. This may be confronting and lead to a more negative attitude towards speech. While scores at pretest and posttest were the same, there were different responses on several items, showing an increased feeling of his speech being regarded 'different' and feeling that 'words don't come out easy'. At follow-up a growth in self-confidence became apparent in items like 'speaking is easy for me' and 'I won't let others speak for me'. The follow-up period, in this case, was a summer break. The boy may have had fewer negative experiences with intelligibility, as he spent more time with family members and had no negative reactions at school or in other social situations. Second, changes in attitude towards own speech may take longer than improvement at the functional level of speech production, as new experiences with improved intelligibility may arise at the end of or even after treatment.

Intelligibility and attitude towards own speech are known to be impacted by inappropriate prosody, consonant and vowel, errors and voicing and nasality errors (Chenausky et al., 2022; Klopfenstein, 2009; McCabe et al., 2014; Hustad, 2012). This suggests that the improvement in intelligibility and attitude in this study may be caused by changes in this type of features related to speech motor

planning and programming. In this study, there were positive changes in these features, which will be discussed next.

Secondary research questions concerned the effect of SMTA on speech motor planning and programming, such as the production of consonants, vowels and clusters in spontaneous speech, picture naming and non-word imitation, as well as DDK measures of speed, accuracy, consistency and fluency. The analysis of the spontaneous speech sample showed progress in the production of consonants, vowels and consonant clusters, suggesting generalization from trained items to spontaneous speech production. Post-hoc analysis showed that consonants were roughly divided into three categories, one category of consonants that was pronounced accurately at pre-test and remained that way, a small category of consonants that showed improvement, and another small category that was persistently difficult throughout the study period. For /r/ and /h/, it is shown in the treatment reports that the boy did produce /r/ and /h/ correctly during practice, but there was no generalization to spontaneous speech in the posttest. Potentially, a larger overall dose for these specific segments, supported with homework, would have resulted in generalization, like the generalization that occurred for cluster production.

Generalization from trained items directly after treatment was found in measures of the CAI. Changes in Z-scores on these measures show that the progress for this boy exceeded growth that would be expected with development. This suggests an effect of treatment that contributed to growth that allowed the boy to (partially) catch-up with his peers.

Improvement was found on measures of the CAI that are related to specific features of CAS, i.e. consistency and the production of clusters in picture naming. At the pre-test the production of consonant clusters in the CCVC syllable structure was below normal limits. There were deletions, resulting in a low score on cluster reductions, just within normal limits. (Note that scores on cluster reduction were inverted, so that a higher score means less cluster reduction.) After treatment scores on both the production of clusters and cluster reduction reached ceiling level. At follow-up the ceiling level performance on cluster reduction was maintained, reflecting that the boy had now fully acquired this syllable structure. A small drop in scores for the production of clusters reflects that some substitutions were present in the realizations at follow-up.

Consistency and the production of clusters also changed in the production of non-words, but this change occurred over the follow-up period, with no improvement directly after treatment. Differences in

scores on tasks with words and non-words might be influenced by auditory skills, which are necessary in the non-word imitation task, but not in picture naming. However, it is unclear why auditory skills would play a role at the post-test and not at follow-up. This would imply an improvement of auditory skills over the follow-up period, for which there are no further indicators. Another explanation might be that new skills need to be automated, and therefore take longer to show up in test results. In that case, it would be expected that all scores for non-word imitation would progress at follow-up, but scores for PCCI and PVC improved directly after treatment. Additional analyses into features of the tasks and items might provide a more robust explanation for the different timing of improvement in consistency and production of clusters in picture naming vs. non-word imitation.

On the DDK task, scores on MRR for both /pata/ and /taka/ sequences showed a clinically relevant decline. This may be influenced by the repeated administrations of the MDT during the treatment phase. The DDK task in the MDT and in the CAI measure DDK in a different way. In weekly testing during treatment, the child was instructed to produce syllables as accurate as possible in the MDT. Producing syllables at optimal speed, as required in the CAI at posttest, would then go against a trained habit. The production of /pataka/, which was impossible for the child at pre-test, did change after treatment. After treatment, production was possible but the score for speed was below normal limits. Similar results were found for /pata/ and /taka/. The habit of optimal accuracy was broken at follow-up, and speed increased again in DDK in CAI for all alternating sequences.

In addition to the pre-test, post-test, and follow-up, a baseline with subsequent weekly testing was conducted of the MDT and a control task. The analysis of different syllable structures and features of speech sounds on the MDT provide detailed insight into speech motor skills at the level of motor planning and programming. Most MDT measures showed a significant trend of improvement over the study period, including measures that relate to specific difficulties in CAS, such as consistency and the production of clusters. The improvement in DDK, a task that places a high demand on speech motor planning and programming, corresponds to the results of the study by Hurkmans (2015).

In this study it was shown that SMTA impacts speech production at the level of speech planning and programming. In a study by Chenausky (2016) minimally verbal children with autism spectrum disorder, some of whom presumably also had CAS improved in speech production after an intervention that used intoning and rhythm. They hypothesized about the effect of unison production and slowed production rate combined with intoning and tapping as facilitators for speech production.

For SMTA, similar mechanisms could explain the effect on speech motor planning and production, but further research is needed to establish which components of SMTA may be considered the working mechanisms in the treatment of CAS.

The non-verbal, cognitive control task that was administered alongside the MDT showed no significant improvement over the study period, suggesting that improvement on the MDT was not caused by developmental progress.

Overall, improvement was obtained on measures that reflect features of CAS and motor planning and programming. These are also measures that reflect more suprasegmental features (cf.co-articulation) of speech. Improvement on these suprasegmental features is in line with both the difficulties this boy experienced in his speech, as well as the rate/rhythm approach of SMTA. The results in this case study may therefore provide a first indication of what the target group for SMTA might be, but further studies are needed to gain more insight into the effect of SMTA in the treatment of different stages and severities of CAS.

Limitations and directions for future research

In this first case study of a series within a proof-of-principle study there were some methodological limitations, especially in the choice of outcome measures. This single-subject design study represents a low level of evidence by itself. However, methodology was optimized, through comparison with a no treatment period and additional control through multiple baseline measurements and a non-speech control task. As the first single subject design into SMTA this study provides sufficient encouragement for follow-up studies.

The measurements in this study were chosen to reflect intelligibility in daily communication, as well as speech motor planning and programming. Objectifying change in intelligibility in daily communication proved to be difficult. In addition to concerns about the scoring of the ICS-Dutch by the father in this specific case, this instrument is vulnerable to bias, as parents are aware of the timing of treatment. Additionally, this instrument provides insight into intelligibility in daily communication, but does not measure whether communicative participation has changed. Changes in communicative participation could be expected when both intelligibility and attitudes towards speech improve, but the current measures did not provide insight into participation. An additional measure, such as Focus on the

Outcomes of Communication Under Six (FOCUS; Thomas- Stonell et al., 2013) might be useful in future studies to get a broader insight into changes in communicative participation after treatment. Results on the CAI show that for some measures, a small change in the raw score may result in a far greater change in Z-score. This is caused by ceiling effects on these measures. Typically developing children at the age of the participant score close to 100% on most measures, reflecting nearly completed motor-speech development (although refinement of these skills continues for longer; Ballard et al., 2012). Therefore, one error affecting the raw score, may cause a large drop in the Z-score for such a measure (or one more item correct may cause a large increase in the Z-score).

The results on the MDT showed that maintenance of the treatment gains mostly occurred for measures that reached proficiency levels of 75% or higher during the treatment phase, such as the correct realization of syllable structures CV and CVC, of place and manner of articulation, and the consistency of productions. This suggests that a child should demonstrate a minimum increase in performance during treatment (in this case a 75% or higher proficiency level) to expect learning (the ability to apply a skill without support; Olswang & Bain, 1994), as reflected by our assessment of maintenance. Additionally, the results on the MDT represent generalization of treatment effects to untreated items. Maintenance of generalized treatment gains holds great potential for improved intelligibility in daily communication. In the present study, the dose of treatment may have been too low to obtain a 75% proficiency level on all measured features. An extended treatment period or increased intensity may be necessary to obtain the proficiency levels required for broader maintenance of improvement.

Measurements were also chosen to objectify speech motor planning and programming. This was specifically measured using the MDT, including consistency, fluency and accuracy. For most measures of the MDT there was a strong dip in the scores towards the end of the treatment period. At this time, the boy turned out to be sick. While results on following test administrations did not completely recover, overall, improvement was still apparent.

Measures of speech planning and programming also provide insight into changes in core features of CAS. Consistency was assessed specifically via the repetition of words and non-words, but also in the repetition of syllables on the MDT. Co-articulatory transitions are assessed via the production of clusters. Prosody was to be assessed through the measure of fluency in the MDT. However, this measure was insufficient in this case to objectify prosody. In the definition of CAS as stated by ASHA

(2007), the realization of lexical and phrasal stress is named as a marker for inappropriate prosody. As the MDT uses non-word syllables, lexical stress is not assessed. Additionally, in this case, scores for fluency approached ceiling levels very early on, as the boy adopted a strategy where he would produce segmentally simplified, inaccurate sequences in a fluent manner. The structure of the task and the strategy adopted by this boy both contribute to a positive, but incorrect reflection of his prosodic skills in the task results.

The assessment of prosody in the evaluation of SMTA in the treatment of CAS is relevant and should be expanded in future studies. First, assessing all three core features of CAS will provide insight into the effect of treatments on these features. This could lead to better choices in treatment planning, choosing a method that is best for a child at a given point in time. Second, SMTA might be especially effective in the treatment of prosody through the support of music. Speech and music are highly related with respect to prosodic features such as frequency (pitch), duration (rhythm) and intensity (dynamics) (Boutsen, 2003; Hurkmans, 2016; Terband et al., 2019). Because of the similarities between music and speech in prosody and the focus of SMTA on the level of speech planning and programming, it is hypothesized that prosody in children with CAS will improve after treatment with SMTA. To evaluate this potential effect, specific measures of prosody at the level of lexical and phrasal stress are needed. Such tasks will be developed to be included in outcome measurements in future studies.

Conclusion

SMTA, a treatment method that combines speech therapy and music therapy, was introduced and it's efficacy in the treatment of CAS is now being evaluated in a single subject design study. The present study shows that speech production in the participant improved after treatment, specifically on tasks that relate to CAS and motor speech planning and programming such as consistency and the production of clusters. While intelligibility improved over the study period, objectifying changes in daily communication proved to be difficult. Additional measures may be necessary to gain more insight into treatment effects at the level of communicative participation.

Overall, the results of this first single subject design study provide sufficient support and important encouragement for further evaluation of SMTA in the treatment of CAS in a proof-of-principle study.

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Data access statement

The data can be obtained by contacting the first author.

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Appendix A: Phonetic repertoire

Appendix B: Medical history of the participant

Appendix C: Trained items

Appendix D: Items and scoring of the Modified Diadochokinesis Test

Figures

Figure 1. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and CIred in Picture Naming. PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct, CCVC = Percentage of Initial Clusters Correct, CIred = cluster reduction. T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 2. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and CIred in Non-word Imitation. PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct, CCVC = Percentage of Initial Clusters Correct, CIred = cluster reduction. T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 3. Raw scores and Z-scores for measures of consistency in word and non-word repetition. WR = word repetition, NWR = Nonword repetition, T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 4. Raw scores and Z-scores for maximum repetition rate in alternating sequences. T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 5. Scores for syllable structure on the MDT. Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

Figure 6. Scores for Consistency, Accuracy and Fluency on the MDT. Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

Figure 7. Scores for Place, Manner and Vowel on the MDT and the control task Figure Weights. Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

Table 1. Structure of a 30 minute treatment session

Phase	Activity	Duration
Introduction	Starting song/ short conversation	3 min
1st exercise	Following protocol in table 2	4 min
2nd exercise	Following protocol in table 2	4 min
Short break	Sing a song or play an instrument	2 min
3rd exercise	Following protocol in table 2	4 min
4th exercise	Following protocol in table 2	4 min
Short break	Sing a song or play an instrument	2 min
5th exercise	Following protocol in table 2	4 min
Conclusion	Make recordings/ sing closing song/ other closing activity	3 min

Note: This structure may vary across children. The time set here per exercise allows for repetition of the entire exercise or practicing a part (word or syllable) of the target item separately before the complete target item.

Table 2. Structure of an SMTA exercise

Step	Participants	Number of repetitions
Introduction of the target word/ sentence	SLP (+ Child)^	1-2 repetitions of target item
Demonstrate target	MT	4-8 repetitions of target item*
Singing	Child + SLP + MT	8-16 repetitions of target item*
Rhythmic chanting	Child + SLP + MT	8-16 repetitions of target item*
Simultaneously speaking	Child + SLP	4-8 repetitions of target item*
Alternately speaking (direct imitation)	Child + SLP	5 repetitions of target item*
Semi-spontaneous speaking (respond to question)	Child + SLP	3 repetitions of target item*

SLP = speech and language pathologist, MT = music therapist. ^At the introduction the child is invited to participate when they are able to repeat the target item correctly. *The number of repetitions is influenced by the length of the target item. A melody usually contains 8 repetitions for words and 4 repetitions for sentences. The melody is sung and chanted at least two times. The numbers here are the minimal number of repetitions that are achieved during one exercise.

884 **Table 3. Schematic representation of outcome measures administered per study phase.**

Pre-test	Baseline (5 times)	Weekly testing	Post-test	Follow-up
ICS-Dutch	MDT	MDT	ICS-Dutch	ICS-Dutch
FAN	Figure Weights	Figure Weights	FAN	FAN
CAI			CAI	CAI
CAT			CAT	CAT
			MDT	MDT
			Figure Weights	Figure Weights

885 ICS-Dutch = Intelligibility in Context Scale – Dutch (McLeod et al., 2012a), FAN = Phonological
886 Analysis for Dutch (Beers, 1995), CAI = Computer Articulation Instrument (Maassen et al., 2019), CAT
887 = Communication Attitude Test (Brutten & Vanryckeghem, 2003), MDT = Modified Diadochokinesis
888 Test (Hurkmans et al., 2012), Figure Weights from the Wechsler Intelligence Scale for Children, Fifth
889 Edition; Dutch Edition (Wechsler, 2018). Full descriptions of these measures are provided under
890 'Outcome measures' in the methods section.
891

892 **Table 4. Scores on the ICS-Dutch as scored by the parents of the participant**.

ICS-Dutch	T1	T2	T3
Father	3.7	3.4	3.9
Mother	3.6	4	4.1
Parents combined	3.6	3.7	4

893 T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment).
894

Table 5. Mean scores of PCCI, PVC and CCVC in spontaneous speech sample

FAN- Dutch	T1	T2	T3
PCCI	85	92	91
PVC	89	97	94
CCVC	40	64	52

PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct, CCVC = Percentage of Initial Clusters Correct. T1 = pre-test, T2 = post-test, T3 = Follow-up (after two months of no treatment).

Table 6. Results for the CAT.

CAT	T1 (Form A)	T2 (Form B)	T3 (Form B)
Raw scores	7	7	3
SD	+ 0.25	+ 0.43	- 0.52

SD = standard deviation, T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment).

Note: A higher score indicates a more negative attitude towards their own speech.

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Speech and music therapy in the treatment of CAS: An introduction and a case study

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The authors report no conflict(s) of interest.

Abstract

Purpose

Speech-Music Therapy for Aphasia (SMTA), a method that combines speech therapy and music therapy, is introduced as a treatment method for childhood apraxia of speech (CAS). SMTA will be evaluated in a proof-of-principle study. The first case study is presented herein.

Method

SMTA was evaluated in a study with a single-subject experimental design comparing 10 weeks of treatment with two months of no treatment. The research protocol included a pre-test, baseline phase, treatment phase, post-test, no-treatment phase and follow-up test. The participant was a five years and eight months old boy with CAS. Outcome measures were selected to reflect both intelligibility in daily communication, as well as features of CAS and speech motor planning and programming.

Results

Results on the Intelligibility in Context Scale-Dutch (ICS-Dutch) and in the analysis of a spontaneous speech sample suggest generalization of treatment effects. Improvements were found in measures that reflect complex speech motor skills, that is, the production of consonant clusters and consistency.

Conclusion

This case study showed that speech production of the participant improved after treatment with SMTA. Although intelligibility as measured with the ICS-Dutch improved over the study period, objectifying changes at the level of intelligibility in daily communication proved to be difficult. Additional measures may be necessary to gain more insight into treatment effects at this level.

Overall, the results of this first case study provide sufficient support and important leads for further evaluation of SMTA in the treatment of CAS in a proof-of-principle study.

Introduction

Childhood Apraxia of Speech (CAS) is a speech sound disorder classified as a subtype of motor speech disorder (Shriberg et al., 2010). In CAS, a core impairment at the level of speech motor planning and programming results in errors in the production of speech sounds and prosody. Inappropriate prosody, inconsistency and disrupted coarticulation are widely described as three key features of CAS (e.g., American Speech-Language-Hearing Association, 2007; Shriberg et al., 1997abc; Terband et al., 2019). Inappropriate prosody, consonant and vowel errors, and voicing and

nasality errors negatively impact intelligibility in CAS (Chenausky et al., 2022; Klopfenstein, 2009; McCabe et al., 2014), which negatively affects functional communication and social participation (Hustad, 2012).

Treatments for CAS address one or more of the three core features of CAS. In Dynamic Temporal and Tactile Cueing (DTTC; Strand, 2020; Strand et al., 2006), a method for children with more severe CAS, all three features are targeted through a focus on movements rather than phonemes, varying prosody and high numbers of repetitions. Rapid Syllable Transition treatment (ReST; Ballard et al., 2010), which is used in older children with less severe CAS, addresses all three features through the focus on sounds, beats and smoothness. Studies report positive outcomes on ratings of production accuracy for DTTC (e.g., Maas et al., 2012; Maas & Farinella, 2012; Strand et al., 2006) and on segmental accuracy and lexical stress for ReST (Ballard et al., 2012; McCabe et al., 2014). While results of these studies for different features of CAS are promising, McCabe and colleagues (2014) showed that the children improved on either lexical stress production or segmental accuracy, but they remained unable to simultaneously produce both correct stress patterns and correct segments. This is known to be a major challenge in developmental speech disorders (Howard, 2007).

Both DTTC and ReST can be described as articulatory-kinematic approaches, using interventions such as visual and tactile cues as well as feedback on the knowledge of performance. ~~They~~These methods also use ~~interventions of the~~some rate/rhythm control ~~type~~strategies, such as reduced speech rate and specific drill of lexical stress. ~~For rate/rhythm control type approaches, interventions are aimed at speech rate, stress and intonation. Examples of~~Other methods, such ~~rate/ rhythm control type approaches are~~as Melodic Intonation Therapy (MIT; Albert et al., 1973; Helfrich-Miller, 1984) and Speech-Music Therapy for Aphasia (SMTA; De Bruijn et al., 2005; Hurkmans et al., 2018~~).~~), are primarily described as rate/rhythm control type approaches. The interventions in these methods are aimed at speech rate, stress, and intonation. MIT and SMTA were originally developed for adults with non-fluent aphasia and Apraxia of Speech (AoS; Hurkmans et al., 2015; Merrett et al., 2014). Both AoS and CAS are described as ~~a disorder~~disorders in the planning and programming of speech movements (American Speech-Language-Hearing Association, 2007; Hurkmans, 2016) and share various characteristics, such as inconsistent errors in the realization of phonemes, syllable segregation, vowel distortions, groping and effect of articulatory complexity (Iuzzini-Seigel & Murray,

2017; Ziegler, 2008). Therefore, rate/rhythm control approaches might be effective in the treatment of CAS.

There is a limited number of studies on the use of rate/rhythm control approaches and music or musical elements in the treatment of CAS (van Tellingen et al., 2022). Four out of eight studies in the systematic review by Van Tellingen et al. (2022) evaluated the use of MIT (Helfrich-Miller, 1994; Krauss & Galloway, 1982; Lagasse, 2012; Martikainen & Korpilahti, 2011). The results ~~in~~of these studies vary and need to be interpreted with caution because the methodological quality of these studies was rated insufficient (van Tellingen et al., 2022).

In the present study, SMTA is evaluated in the treatment of CAS in the first case of a series in a single subject experimental design. The background and protocol for SMTA are introduced in the next section.

SMTA

SMTA is a combination of speech therapy and music therapy in which a speech language pathologist (SLP) and a music therapist (MT) provide the treatment simultaneously. It is used in clinical practice with children from three years onwards with motor speech disorders, including (suspected) CAS. This method uses musical parameters that support the prosody of speech on word, phrase, and sentence levels and facilitate the sequencing and timing of speech movements. The musical compositions are tailored to ~~the~~ individual needs, as the music therapist composes melodies to support the functionally relevant speech targets.

There are two lines of treatment: a speech therapy line and a music therapy line, that are conducted simultaneously. Speech therapy includes three levels: (1) syllables, (2) words and (3) sentences. These levels allow for a focus on movements, rather than individual speech sounds. As an exception, vowels may be practiced in isolation at the first level. Target items on the word and sentence levels are designed both to fit the speech targets based on speech assessment, as well as to be functionally relevant (and therefore motivating) for the individual child in daily communication. For example, when a child has difficulty producing consonant clusters, and their brother's name is 'Steven', this could make this name an excellent target item. Items may be both personal, such as names of family members, and more formulaic ~~items~~, such as 'thank you'. Music therapy follows a structured procedure that starts with singing, followed by rhythmical chanting and speaking, which is derived from

MIT. In SMTA, the final step of speaking is divided into smaller steps, including simultaneous speaking, alternating and semi-spontaneous speech. MIT uses rhythm and melody to simplify and exaggerate prosody, limiting melody in an alternation of a limited number of pitches (Sparks, 1981). The exercises in SMTA are designed to musically support natural speech, using the musical parameters melody, rhythm, meter, tempo and dynamics. For each target item a new melody is composed to support the prosodic features of the spoken utterance. This allows for selection of targets that are specifically tailored to the communicative needs and interests, speech sound inventory, and speech motor processes of the child. During an exercise, musical parameters may be used to adjust the exercise as needed. Variations in tempo, for example, may increase or decrease the difficulty of the exercise (De Bruijn et al., 2005; Hurkmans et al., 2018). SMTA has been shown to be an effective treatment method for Dutch adults with AoS and aphasia in a proof of principle study with five speakers with aphasia and AoS (Hurkmans et al., 2015). In ~~this~~that study, intelligibility of verbal communication in daily life improved, as well as articulation.

SMTA is originally based on various similarities between language and music, such as shared hierarchical structures (Hurkmans, 2016; Patel, 2003; Peretz & Zatorre, 2003) and shared neural processing (e.g. Brown et al., 2006; see Hurkmans ~~(~~[2016~~)~~] for further discussion). SMTA has been shown to improve speech production at the level of motor planning and programming (Hurkmans, 2015). To provide the rationale ~~of~~for SMTA in the treatment of CAS, three theoretical frameworks on music and speech and the potential working mechanisms of music in the treatment of speech will be discussed~~:~~: (1) similarities between and overlap in the processing of speech and music, (2) overlap in prosodic features in music and speech, and (3) mechanisms of music with regards to motivation and mood.

One of the original fundamental ideas for SMTA is the overlap in neural processing for language and music, which can be expanded to speech. Fujii and Wan (2014) showed that an overlap in neural processing of rhythm in music and speech, combined with synchronization and entrainment to a pulse, explains how rhythm supports the recovery of speech production. Overlap between music and speech can also be found in prosody, which is characteristic for music and speech (e.g., ~~(~~Boutsen, 2003). Both include highly related features of sound such as melody and pitch, rhythm and duration, and dynamics and intensity (Hurkmans, 2016). Pitch, duration, and intensity are described as the features that combine to express stress in many languages (Terband et al., 2019). Through the overlap in

features music can be used to support speech prosody (Hurkmans et al., 2015). The third theoretical framework concerns the positive effects of music on mood and motivation in speech-language interventions, which have been summarized by Merrett, Peretz and Wilson (2014) as one of the possible working mechanisms of MIT. In short, music is believed to have a positive effect on mood and motivation, which may contribute to the effect of interventions that utilize musical elements. Together, these frameworks provide insight ~~in~~into the potential mechanisms that contribute to the ~~effect~~effects of SMTA.

SMTA encompasses all principles of motor learning that are recommended in the treatment of CAS (Maas et al., 2014; Murray et al., 2014; Strand, 2020). The use of music in SMTA allows for a high number of trials per session, which is crucial for motor learning (Maas et al., 2014; Strand, 2020), as singing (including a minimum of 20 trials per exercise) is regarded as more pleasant than realizing a high number of trials in a drill-type exercise. Usually up to five different exercises are conducted during a treatment session, ~~alternated~~alternating with small musical activities, such as singing a song or playing an instrument. Children are given autonomy within the session as they are invited to select the targets they want to practice and choose the musical activities they wish to engage in. This autonomy and the highly relevant target items can both contribute to increased motivation (Strand, 2020; Wulf et al., 2018). During an exercise, verbal feedback is kept to a minimum, to avoid disruptions of the flow of the exercise. Non-verbal knowledge of performance feedback is provided by focusing the child's attention on the provided oral example in which the speech therapist may emphasize a specific movement. Feedback ~~regarding~~in the form of knowledge of results is also provided non-verbally and focusses on accurate realizations of the target word. Placing the child's attention on accurate realizations raises their expectancies of their own ability. Before and at the end of an exercise, the functional use of the target is emphasized, ~~as a way~~ to direct the attention to an external focus on results. This focus on feedback at the level of knowledge of results is recommended in the treatment of children with CAS to stimulate learning and retention (Strand, 2020) and has consistently been shown to enhance learning regardless of task, age, skill level, or (dis)ability (Wulf et al., 2018).

Treatment protocol

SMTA is provided by trained SLPs and MTs. When a child is referred for SMTA, the SLP formulates target items with the child and/ or their parents or caregivers, depending on the age of the child. These

target items are at the word and/ or sentence level and are selected to be both functionally relevant for the child and relevant for the speech targets that are formulated based on speech assessment. Target items ~~on~~at the syllable level may be added for consonants or clusters that are still difficult for the child. Ideally, these targets at the syllable level will subsequently be used in targets at word and/ or sentence levels.

The music therapist composes new melodies that support the natural melody, rhythm, and prosody of the target items. To this end, the music therapist uses melody, meter, rhythm, tempo, and dynamics to compose a melody that is close to the spoken prosody of the target item. This implies that musical features such as complex melodic structures, large intervals and syncopation should be avoided (for Western languages). The musical parameters can also be used to influence the difficulty of the exercise, e.g~~.~~., for meter, a 6/8 beat elicits fluency more than a 4/4 beat. The composed melody consists of repetitions of the target item. The number of repetitions varies with the length of the target item, e.g~~.~~., four repetitions for a sentence or eight repetitions for a word. All melodies are new and specifically composed for the target item, as famous or ~~previous~~previously used melodies will elicit the words and sentences that go with these melodies.

During a therapy session of 30 minutes up to five target items will each be practiced in a fixed structure (see Table 1 and 2). The child is usually seated opposite of the SLP, for visual assistance, e.g~~.~~., the oral example that the SLP provides. The MT is seated beside the child and SLP, creating a triangle shaped setup. The introduction of the target item by the SLP includes naming the target clearly. The introduction may be supported by an object, photograph, or picture of the target item. This is followed by a demonstration of the target item by the MT. Directly ~~hereafter~~thereafter, the child and the SLP join in with the MT and sing the melody, usually two times, but more repetitions can be added if deemed necessary. The next step is rhythmic chanting. In this phase the melody fades out and the musical support is reduced to rhythmic assistance. The child and SLP can join in with the rhythmic support by tapping with the hand or foot, but this is not required. The choice to do this should be made based on ~~the basis of~~ what is helpful and not distracting for the child in their efforts to produce the target item. After rhythmic chanting the rhythmic support is removed and the SLP and the child simultaneously produce the target item repeatedly. Then the SLP introduces turn-taking (direct imitation) with a hand gesture, signaling when the SLP will speak and when the child is invited to speak. Finally, the SLP poses a question that will elicit the target item. In this step, any visual support

used in ~~introduction~~the introductory phase may be used to repeatedly elicit the target item. During the exercises and between the phases, verbal feedback is to be kept to a minimum. Feedback may be non-verbal, with facial expression or small gestures, but interruptions of the flow of the exercise should be avoided.

Changes may be necessary during the exercise, such as a change in tempo or an extra repetition of singing. Ideally, the SLP and MT develop a cooperation so that these changes can be made during the exercise by non-verbal cues to one another, without disrupting the flow of the exercise.

Most children enjoy a break between exercises with singing a favorite song or playing an instrument for a short amount of time.

Insert table 1 and 2 around here.

When a child produces a target item correctly at semi-spontaneous (elicited) speaking during therapy, recordings of the complete exercise, can be made for practice at home. While practice at home with these recordings lacks the opportunity to provide interventions during the exercise, it does create opportunities for increasing treatment dose. Practicing at home might also contribute to the transfer of the target items to spontaneous speech in daily communication outside the treatment setting as realization of a target item in the pragmatically intended context provides a greater experience of success.

Clinical experiences with SMTA in the treatment of CAS are positive, but up to now there were no efficacy studies of SMTA in the treatment of CAS. This study represents the first single subject design study ~~on~~into the effectiveness of SMTA in the treatment of CAS. ~~The purpose of this study was to evaluate the efficacy of SMTA in the treatment of a child with CAS.~~ The main research question was whether intelligibility in daily communication improves after treatment with SMTA. Secondary research questions focused on the effect of SMTA on the production of consonants, vowels and clusters in spontaneous speech, picture naming and non-word imitation, as well as measures of speed, accuracy, consistency and fluency in a diadochokinesis task (DDK).

Case study

Method

SMTA was evaluated in a study with a single-subject experimental design comparing 10 weeks of treatment with two months of no treatment focusing on speech production (at the level of phonological encoding and speech planning and programming). The research protocol included a pre-test, baseline phase, treatment phase, post-test, no-treatment phase and follow-up test. The protocol was approved by the research ethics committee at the University of Groningen (ref.nr. 77088008). Parents gave written informed consent prior to participation in the study. Treatment was provided at a rehabilitation center by an SLP and MT who were both trained and experienced in providing SMTA. They followed the protocol for SMTA described in the introduction. Test administrations were conducted by another SLP, who was unaware of treatment progress. All tests at all timepoints were administrated by the same SLP, at the same location, using the same equipment for test administration and recording. Recordings of the test administrations were scored by the first author, who was blinded to the order of the recordings during scoring. After scoring was completed, results were matched to their date with the key provided by the SLP who conducted the test administrations.

Participant

The participant was a 5;~~8y~~ 8 year-old Dutch-speaking boy with CAS. The diagnosis of CAS was confirmed following the protocol ~~by~~of Iuzzini-Seigel and Murray (2017), assessing several features of CAS in various speech tasks. The boy presented with inconsistent speech ~~in~~on a word and non-word repetition task and in spontaneous speech. Additional features included increasing problems with increasing complexity or length which was shown in spontaneous speech, picture naming, non-word repetition and DDK. Syllable segmentation, groping and elongation of initial consonants were observed throughout tasks. There were some consonant deletions and substitutions. In the phonological analysis of ~~his~~the child's spontaneous speech, the consonants /h/ and /r/ were produced ~~accurate~~accurately less than 50% of occurrences in initial position. Additionally, the consonants /s/, /d/ and /x/ (velar fricative) were produced correctly between 75% and 100% of the time in initial position. For /d/, there were several deletions in multi-syllabic words. In monosyllabic contexts, /d/ was realized accurately. In word-final position, the consonant /m/ was produced accurately in 33% of occurrences, and /l/, /k/, /n/ and /t/ were produced accurately in between 78% and 94% of occurrences. Other consonants were produced correctly in 100% of occurrences, both in initial and final position. A full

overview of Dutch phonemes and this boy's phoneme acquisition is presented in appendix A. The participantsparticipant had voicing difficulties, leading to whispering partial and complete utterances across speech tasks. Intelligibility was negatively influenced by suprasegmental features, such as the difficulties with voicing, dysfluency, low speech rate and increasing difficulties with increasing length and/or complexity. Segmental errors impacted intelligibility to a lesser extent, with the absence of /h/ and /r/ in his speech being striking, but also consistent. He showed awareness of his speech problems and a lack of self-confidence while speaking.

The medical history of the boy is described in detail in appendix B. His medical history included gross motor difficulties, including delayed development of walking. His fine motor skills were age appropriate and after physical therapy, his gross motor skills were age appropriate when he was 3;2 (years; months) old. He had persistent colds and tonsil issues around the age of 18 months, resulting in the clipping of his tonsils and placement of tubes in his ears around his second birthday. His hearing was within normal limits when measured at the age of 20 months and again at age 3;7.

His speech and language were assessed multiple times from the age of 2 to track progress and treatment effecteffects. Word and sentence comprehension were within normal limits. Productive vocabulary showed delay, but was within normal limits from the age of 3. Sentence production was below normal limits. Treatment was focused on increasing speech production, through targeting speech sounds and syllables. Non-verbal psychological assessment was conducted at the audiological center when the boy was 3;9, showing a normal non-verbal psychological development. The boy was placed atin a specialized early education group, focusing on speech and language when he was 3;9. When he was 4;5, he went to school (which corresponds to preschool in the USA). He continued speech therapy in private practice. While there was progress in his phonological development, features of CAS became more apparent. Therefore, he was referred to the rehabilitation center for further assessment of suspected CAS and treatment at the age of 5;3.

Intervention

After the pretest, there was a two-week baseline phase. In this period five baseline measures (see below for a description of the measures) were taken. After the baseline phase treatment started, consisting of two 30-minute sessions of SMTA per week for 10 weeks, with additional homework using recordings of the targets that havehad been realized successfully during treatment. 10 target items on

the sentence level were drawn up by the speech therapist and parents together. These items, which are presented in appendix C, were both functionally relevant to the child as well as fitting with the outcomes of his speech assessment. Items were drawn up to target both the persistent segmental errors (/r/ and /h/) and the suprasegmental features, through choosing items at the sentence level, with multisyllabic words, and including numerous consonant clusters. Items were introduced over the treatment period as the SLP and MT saw fit.

A posttest assessment was administered after the treatment phase and a ~~posttest measurement~~follow-up assessment was ~~conducted, followed by~~administered after a two-month no-treatment phase~~ of two months. Finally, a follow-up measure was conducted.~~

Outcome measures

A schematic overview of the outcome measures and timing of administration is presented in table 3. The primary outcome measure was chosen to reflect intelligibility in daily communication, in line with the core objective of speech therapy, to support the child in communicative participation in society (Hustad, 2012). The selected measure was the Intelligibility in Context Scale – Dutch (ICS-Dutch; McLeod et al., 2012a). In this questionnaire, parents rate the intelligibility of their child in contact with various communication partners, such as family members, peers, teachers, and strangers on a five-point scale. Reliability and validity for this instrument was assessed for the original English version (McLeod et al., 2012b) and the Dutch version used in the current study (McLeod, 2020; Van Doornik et al., 2018) and found to be adequate. Both parents filled out the ICS-Dutch independent of one another.

Further outcome measures were selected to reflect speech motor abilities in various tasks, such as spontaneous speech, picture naming, non-word repetition and DDK.

A speech sample was collected and analyzed using the Phonological Analyses for Dutch (Fonologische Analyse voor het Nederlands (FAN); Beers, 1995; Beers & Masereeuw, 2022). The sample was elicited through a series of standardized questions on topics such as school, hobbies, and vacation. The first 100 unique words in the sample were transcribed and analyzed. In this task Percentage Consonants Correct in syllable initial position (PCCI) and Percentage Vowels Correct (PVC) were calculated, as well as percentage of clusters correct in syllable initial position (CCVC).

The Computer Articulation Instrument (CAI; Maassen et al., 2019) was used to assess speech in specific tasks. The CAI consists of the subtests Picture Naming, ~~Nonword~~Non-word Imitation, Word- and ~~nonwordrepetition~~Non-Word Repetition and DDK. For Picture Naming and Non-word Imitation PCCI, PVC and CCVC were calculated. Additionally, the occurrence of ~~Clusterreduction~~cluster reduction (Clred) was calculated. For Word- and ~~nonwordrepetition~~non-wordrepetition consistency was calculated and for DDK the measure was Maximum Repetition Rate (MRR). The CAI ~~has~~is norm-referenced and has been shown to have sufficient to good reliability and validity for the assessment of speech development in Dutch children ages 2-7 (van Haaften et al., 2019). The CAI norms are divided into age groups that span four months for the younger groups and six months for the two oldest groups. For this study, raw scores for the pre-test and post-test were compared with the norms for children aged 5;8-5;11 and raw scores for follow-up were compared to the norms for children aged 6;0-6;5 (van Haaften et al., 2019).

The Communication Attitude Test (CAT; Brutten, 1984; Brutten & Vanryckeghem, 2003) was used to assess the attitude of the child towards their own speech. In this norm-referenced instrument, children respond to statements about their speech with true or false. A higher score indicates a more negative attitude towards their own speech. The CAT was originally developed for children who stutter. The ~~instruments~~instrument's reliability and validity have been studied in children who stutter and were sufficient (Vanryckeghem & Brutten, 1992).

The before-mentioned tasks were assessed at pre-test, post-test and follow-up. Additionally, the Modified Diadochokinesis Test (MDT; Hurkmans et al., 2012) was used to ~~assess~~assess speech motor planning and programming and establish a baseline and track treatment progress through weekly assessment. The MDT is a qualitative assessment of DDK. It consists of items ~~in~~of CV, CVC, CVCC and CCVC structure, in which the vowel, or place or manner of the consonant varies (see appendix D for the items). The child is instructed to repeat each item five times, as ~~accurate~~accurately as possible. Responses are scored for accuracy, fluency and consistency. Speech rate is not assessed. The reliability and validity of the MDT were assessed for use with adults with AoS and were sufficient for this group (Hurkmans et al., 2012). There are no records of the reliability and validity of the MDT for use with children with CAS.

A non-verbal control task was used to control for developmental progress. This was the task Figure Weights from the Wechsler Intelligence Scale for Children, Fifth Dutch edition (WISC-V-NL; Wechsler,

2018). The Figure Weights task was used in the baseline ~~and~~, weekly testing, and follow-up testing, alongside the MDT. Reliability and validity of the WISC-V-NL are adequate (Wechsler, 2018).

Insert table 3 around here

Analysis

Kendall's tau test was used to test for change on the MDT measures CV, CVC, CCVC, CVCC, accuracy, consistency, fluency, place, manner and vowel. The level of significance was set at $p<0.05$. Kendall's tau test was also used to analyze ~~the~~ changes in the non-verbal control task Figure Weights. The CAI and CAT ~~are norm-referenced. These~~ norms were used to describe changes on these measures. ~~Norm groups in CAI are divided into age groups that span four months for the younger groups and six months for the two oldest groups. For this study this means that raw scores for pre-test and post-test were compared with the norm group for children aged 5;8-5;11 and raw scores for follow-up were compared to the norm group for children aged 6;0-6;5 (van Haaften et al., 2019).~~

Results

The participant in this study received 20 30-minute SMTA sessions, over a period of 11 weeks instead of 10, due to one week of illness during the treatment period. The number of ~~(partial) items~~items (both full items at the sentence level and parts of items at the word level) that was trained in each session varied from four to seven. The SLP and MT implemented ~~repetition~~repetitions of items and breaks as needed, resulting in different numbers of items practiced across sessions. In one session there were four trained items, in ~~12~~twelve sessions there were five, in five sessions there were ~~6~~six trained items and in two sessions there were seven. Each week, the items trained in the first session were repeated in the second. The first week, the more personally ~~motivated~~motivating item with the name of his stuffed animal was trained, building from practicing words to the full sentence. In the second and third week, items with the persistently difficult /r/ and /h/ were practiced at the word and sentence level. In the fourth through sixth week, ~~the~~ items with consonant clusters were trained at the syllable, word and ~~sentences~~sentence levels. From week seven on, focus was placed on ~~the~~ multisyllabic words, as well as consonant clusters, building up to sentence level for items containing such words. Nine out of ten of the formulated targets at sentence level were trained. The boy was able to produce the fourth item

correctly upon first request during treatment~~,~~. Therefore this item was not further trained. For all trained items it was necessary to practice words or syllables separately. This concerned words with consonants that were difficult for this boy, such as /h/ and /r/. Several consonant clusters were trained at the syllable level. Musical interventions were used to support speech production, such as decreased tempo for multisyllabic words and upbeats for the production of clusters. Speech therapy interventions included verbal instructions and oral examples. For example, the boy produced an interdental ~~/~~[n~~/~~] as a substitute for /ŋ/. Verbal instruction and oral ~~example~~examples to keep his mouth wide open ~~in~~for the syllable /aŋ/ supported the production of this syllable correctly. This was subsequently integrated ~~in~~into the word 'belangrijk' (*important*~~) and~~); /ŋ/ was easier achieved in the item 'springen' (*jump*) that was trained later on. ~~Besides~~In addition to instances of feedback at the level of knowledge of performance, feedback on knowledge of results was provided through non-verbal signs. These included signals to increase attention and effort, as well as reinforcements for adequate productions. Frequency of feedback was decreased with increasing adequate productions. Homework was provided, without fixed guidelines or expectations. Recordings of items that the boy could produce correctly during practice were sent to parents through a secure e-health application. Parents reported ~~to have~~having had limited opportunities for practice at home. The participant received no ~~(~~other speech~~ therapy)~~ treatment at all during the second phase of this study, as this coincided with a summer break.

Results for intelligibility as measured with the ICS-Dutch, are presented in table 4. Combining the scores of both parents, the results suggested some improvement over the study period. Scores of the father and mother individually show different patterns.

Insert table 4 around here.

The analysis of a sample of spontaneous speech with the FAN suggested improvement ~~for~~on PCCI over the treatment period, as presented in table 5. Improvement was found for /x/ and /s/, which were realized accurate in 100% of occurrences after treatment, which was maintained at follow-up. The realization of /d/ improved after treatment and improved further over the follow-up period. For the consonant /r/ there was some improvement after treatment, which was not maintained at follow-up. Improvement ~~for~~on PVC and the production of initial clusters ~~that~~, which was obtained after treatment, was not maintained at follow-up.

Insert table 5 around here.

Tasks and measures ~~of~~from the CAI showed varying results. The CAI does not provide critical differences, therefore a change of Z > + 0.5 was set as the norm for clinically relevant change. Z-scores were calculated using the means and SDs published by Van Haaften et al. (2019). In Picture Naming, presented in figure 1, there was clinically relevant change for PCCI over the study period (Z + 0.51 at follow-up compared to pre-test), but no clinically relevant changes for PVC (Z + 0.42 over treatment period). The production of initial clusters in Picture Naming improved after treatment (Z + 5.46), and the gains were mostly maintained at follow-up (Z + 4.29). Results for cluster reductions in the same task, for which scores were inverted to reflect that a higher score means ~~less~~fewer cluster reductions, showed clinically relevant change over the treatment period, reaching ceiling level after treatment (Z + 0.97), which was maintained at follow-up (Z + 1.16). ~~In~~On Non-Word Imitation, presented in figure 2, results for PCCI showed clinically relevant change after treatment (Z + 1.22), but this was not maintained at follow-up (Z + 0.16). Results for PVC showed an increase from below average to within normal limits directly after treatment (Z + 0.85), but this improvement was not maintained at follow-up (Z ~~-~~– 1.0). The production of initial clusters in Non-Word Imitation did not change directly after therapy, but did improve at follow-up (Z + 1.34). Results for cluster reduction in this task showed clinically relevant change, reaching ceiling level after treatment (Z + 2.8), which was maintained at follow-up (Z + 2.83). Results for consistency are presented in figure 3. Word consistency increased directly after treatment (Z + 0.80), but this improvement was not maintained at follow-up. Non-word consistency did not increase directly after treatment, but improvement was apparent at follow-up (Z + 1.71). The DDK ~~of~~task from the CAI, which measures maximum repetition rate (MRR; syllables per second) showed no change for the sequential items /pa/, /ta/ and /ka/. Results for the alternating items are presented in figure 4. For the items /pata/ and /taka/, results were lower directly after treatment (Z – 0.48 and Z – 2.42) and at follow-up (Z – 0.16 and Z – 0.69). For /pataka/ the scores improved after treatment (Z + 3.59) from not being able to perform this sequence at pre-test to within normal limits at follow-up (Z + 4.21).

Insert figures 1,2, 3 and 4 around here

Results for communication attitude as measured with the CAT are presented in table 6. There was no change in scores directly after treatment. At follow-up scores were lower, showing a more positive attitude towards speech.

Insert table 6 around here

Results on several measures of the MDT showed change after treatment and over the study period. For syllable structure, there was a significant gradual improvement on CV (Kendall $\tau = 0.633$, $p < .01$) and CCVC (Kendall $\tau = 0.396$, $p < .05$) structures as shown in figure 5. Results for the structures CVC (Kendall $\tau = 0.290$, $p > .05$) and CVCC (Kendall $\tau = 0.132$, $p > .05$) showed no significant change over the study period. Significant gradual improvement was found ~~in~~for the measures of accuracy (Kendall $\tau = 0.433$, $p < .05$) and consistency (Kendall $\tau = 0.447$, $p < .05$), but not for fluency (Kendall $\tau = 0.211$, $p > .05$) as shown in figure 6. Figure 7 shows significant gradual improvement for the measures place (Kendall $\tau = 0.513$, $p < .01$), manner (Kendall $\tau = 0656$, $p < .01$) and vowel (Kendall $\tau = 0.356$, $p < .05$). ~~On~~For the scores on the non-verbal control task Figure Weights, which is also shown in figure 7, there ~~is~~was no significant improvement over the study period (Kendall $\tau = -0.210$, $p > .05$).

Insert figure 5, 6 and 7 around here

Discussion

In this article we introduced Speech-Music Therapy for Aphasia (SMTA) as a new method in the treatment of childhood apraxia of speech (CAS). SMTA combines speech therapy and music therapy and is designed to support speech production at the level of motor planning and programming. Its potential for the treatment of CAS is supported by evidence of its effectiveness in adults with Apraxia of Speech (AoS; Hurkmans et al., 2015) as well as theoretical frameworks ~~on~~of the neural processing of rhythm in speech and music (Fujii & Wan, 2014), similarities between speech and music at the level of prosody (Hurkmans, 2016; Terband et al., 2019), and the positive effects of music on mood and motivation (Merrett et al., 2014). As a first investigation of this potential, we ~~subsequently~~ evaluated the effectiveness of SMTA in the treatment of CAS in a multiple baseline single subject design. First

and foremost, this study showed that SMTA can be administered to five- to six-year-old children with CAS. Furthermore, the treatment yielded positive outcomes in speech production and intelligibility for the participant in this case study.

The study was designed to evaluate SMTA on a range of outcome measures, including intelligibility in daily communication and communication attitude, as well as a variety of measures of speech motor planning and programming in specific speech ~~of speech~~ motor tasks. Tasks included picture naming, non-word imitation, word- and non-~~word~~repetitionword repetition, and DDK, with outcome measures such as percentage of initial consonants correct, production of clusters, consistency and fluency. Overall results showed progress ~~in~~on measures of speech motor planning and programming occurring directly after treatment. Improvement ~~of~~on intelligibility and communication attitude was mostly obtained over the entire study period and became apparent at follow-up.

As the goal of treatment in clinical practice lies in optimizing communication in daily ~~live~~life, we chose intelligibility in daily communication as the main outcome measure. In the scores of both parents on the ICS-Dutch combined, improvement was found at follow-up, but no effect was found directly after treatment. Examining the individual data of the two parents revealed that the father scored lower intelligibility directly after treatment. A possible explanation for this might be an increased awareness of his ~~sons~~son's speech problems through the pre-test and treatment ~~phase~~phases. Participation in the study and the parental questionnaires filled out at the beginning of the study might have drawn attention and raised awareness of the speech problems, leading to lower scores directly after treatment.

A similar trend was found in the scores of the CAT, with improvement in the boy's attitudes towards his own speech apparent at follow-up, but not directly after treatment. This was observed in both the results on the CAT and clinical observations by the SLP who conducted the test administrations ~~and~~as well as by the first author who judged and scored the video-recordings of spontaneous speech and test administrations. Clinical observations included changes in posture, communicative initiative and speaking more freely. Two underlying mechanisms may have contributed to the pattern of improvement at follow-up rather than directly after treatment. First, during treatment, focus is put on speech and the difficulties with speech. This may be confronting and lead to a more negative attitude towards speech. While scores at pretest and posttest were the same, there were different responses on several items, showing an increased feeling of his speech being regarded 'different' and feeling

that ‘words don’t come out easy’. At follow-up a growth in self-confidence became apparent in items like ‘speaking is easy for me’ and ‘I won’t let others speak for me’. The follow-up period, in this case, was a summer break. The boy may have had ~~less~~fewer negative experiences with intelligibility, as he spent more time with family members and had no negative reactions at school or in other social situations. Second, changes in attitude towards own speech may take longer than improvement at the functional level of speech production, as new experiences with improved intelligibility may arise at the end of or even after treatment.

Intelligibility and attitude ~~in CAS~~towards own speech are known to be impacted by inappropriate prosody, consonant and vowel, errors and voicing and nasality errors (Chenausky et al., 2022; Klopfenstein, 2009; McCabe et al., 2014; Hustad, 2012). This suggests that the improvement in intelligibility and attitude in this study may be caused by changes in this type of features related to speech motor planning and programming. In this study, there were positive changes ~~in~~ in these features, which will be discussed next.

Secondary research questions concerned the effect of SMTA on speech motor planning and programming, such as the production of consonants, vowels and clusters in spontaneous speech, picture naming and non-word imitation, as well as DDK measures of speed, accuracy, consistency and fluency. The analysis of the spontaneous speech sample showed progress in the production of consonants, vowels and consonant clusters, suggesting generalization from trained items to spontaneous speech production. Post-hoc analysis showed that consonants were roughly divided into ~~two~~three categories, one category of consonants that was pronounced accurately at pre-test and remained that way, ~~and a~~a small category of consonants that showed improvement, and another small category that was persistently difficult throughout the study period. For /r/ and /h/, it is shown in the treatment reports that the boy did produce /r/ and /h/ ~~correct~~correctly during practice, but there was no generalization to spontaneous speech in the posttest. Potentially, a larger overall dose for these specific segments, supported with homework, would ~~result~~have resulted in generalization, like the generalization that occurred for cluster production.

Generalization from trained items directly after treatment was found in measures of the CAI. Changes in Z-scores on these measures show that the progress for this boy exceeded growth that would be expected with development. This suggests an effect of treatment that contributed to growth that allowed the boy to (partially) catch-up with his peers.

Improvement was found on measures of the CAI that are related to specific features of CAS, i.e. consistency and the production of clusters in picture naming. At the pre-test the production of consonant clusters in the CCVC syllable structure was below normal limits. There were deletions, resulting in a low score on cluster reductions, just within normal limits. (Note that scores on cluster reduction were inverted, so that a higher score means less cluster reduction.) After treatment scores on both the production of clusters and cluster reduction reached ceiling level ~~(scores on cluster reduction where inverted, so that a higher score means less cluster reduction).~~. At follow-up the ceiling level ~~of~~performance on cluster reduction was maintained, reflecting that the boy had now fully acquired this syllable structure. A small drop in scores for the production of clusters reflects that some substitutions were present in the realizations at follow-up.

Consistency and the production of clusters also changed in the production of non-words, but this change occurred over the follow-up period, with no improvement directly after treatment. Differences in scores on tasks with words and non-words might be influenced by auditory skills, which are necessary in the non-word imitation task, but not in picture naming. However, it is unclear why auditory skills would play a role at the post-test and not at follow-up. This would imply an improvement of auditory skills over the follow-up period, for which there are no further indicators. Another explanation might be that new skills need to be automated, and therefore take longer to show up in test results. In that case, it would be expected that all scores for ~~nonword~~non-word imitation would progress at follow-up, but scores for PCCI and PVC improved directly after treatment. Additional analyses into features of the tasks and items might provide a more robust explanation for the different timing of improvement in consistency and production of clusters in picture naming ~~and~~vs. non-word imitation.

~~In~~On the DDK task, scores on MRR for both /pata/ and /taka/ sequences showed a clinically relevant decline. This may be influenced by the repeated administrations of the MDT during the treatment phase. The DDK task in the MDT and in the CAI measure DDK in a different way. In weekly testing during treatment, the child ~~is~~was instructed to produce syllables as accurate as possible in the MDT. Producing syllables at optimal speed, as required in the CAI at posttest, would then go against a trained habit. The production of /pataka~~/~~/, which was impossible for the child at pre-test, did change after treatment~~, as this was not possible at pre-test~~. After treatment, production was possible but the score for speed was below normal limits. Similar results were found for /pata/ and /taka/. The habit of

optimal accuracy was broken at follow-up, and speed increased again in DDK in CAI for all alternating sequences.

~~Apart from~~In addition to the pre-test, post-test, and follow-up, a baseline with subsequent weekly testing was conducted ~~with~~of the MDT and a control task. The analysis of different syllable structures and features of speech sounds ~~in~~on the MDT provide detailed insight ~~in~~into speech motor skills at the level of motor planning and programming. Most MDT measures ~~show~~showed a significant trend of improvement over the study period, including measures that relate to specific difficulties in CAS, such as consistency and the production of clusters. The improvement in DDK, a task that places a high demand on speech motor planning and programming, corresponds to the results of the study by Hurkmans (2015).

In this study it was shown that SMTA impacts speech production at the level of speech planning and programming. In a study by Chenausky (2016) minimally verbal children with autism spectrum disorder, some of ~~which~~whom presumably also had CAS improved in speech production after an intervention that ~~uses~~used intoning and rhythm. They hypothesized ~~on~~about the effect of unison production and slowed production rate combined with intoning and tapping as facilitators for speech production. For SMTA, similar mechanisms could explain the effect on speech motor planning and production, but further research is needed to establish which components of SMTA may be considered the working mechanisms in the treatment of CAS.

The non-verbal, cognitive control task that was administered alongside the MDT showed no significant improvement over the study period, suggesting that improvement on the MDT was not caused by developmental progress.

Overall, improvement was obtained on measures that reflect features of CAS and motor planning and programming. These are also measures that reflect more suprasegmental features (cf.co-articulation) of speech. Improvement on these suprasegmental features is in line with both the difficulties this boy experienced in his speech, as well as the rate/rhythm ~~type~~ approach of SMTA. The results in this case study may therefore provide a first indication of what the target group ~~of~~for SMTA might be, but further studies are needed to gain more insight ~~in~~into the effect of SMTA in the treatment of different stages and severities of CAS.

Limitations and directions for future research

In this first case study of a series ~~for~~within a proof-of-principle study there were some methodological limitations, especially in the choice of outcome measures. This single-subject design study represents a low level of evidence ~~on~~by itself. However, methodology was optimized, through comparison with a no treatment period and additional control through multiple baseline measurements and a non-speech control task. As the first single subject design into SMTA this study provides sufficient ~~leads~~encouragement for ~~following~~follow-up studies.

The measurements in this study were chosen to reflect ~~both~~ intelligibility in daily communication, as well as speech motor planning and programming. Objectifying change in intelligibility in daily communication proved to be difficult. ~~Besides the issues with~~In addition to concerns about the scoring of the ICS-Dutch by the father in this specific case, this instrument is vulnerable to bias, as parents are aware of the timing of treatment. Additionally, this instrument provides insight ~~in~~into intelligibility in daily communication, but does not measure whether communicative participation ~~changes~~has changed. Changes in communicative participation could be expected when both intelligibility and attitudes towards speech improve, but the current measures did not provide insight ~~in~~into participation. An additional measure, such as Focus on the Outcomes of Communication Under Six (FOCUS; Thomas- Stonell et al., 2013) might be useful in future studies to get a broader insight ~~in~~into changes in communicative participation after treatment.

Results on the CAI show that for some measures, a small change in the raw score may result in a far greater change in Z-score. This is caused by ceiling effects on these measures. Typically developing children ~~in~~at the age of the participant score close to 100% on most measures, reflecting ~~a~~ nearly completed motor-speech development (although refinement of these skills continues for longer; Ballard et al., 2012). Therefore, one error ~~in~~affecting the raw ~~scores~~score, may cause a large drop in the Z-score for such a measure (or one more item correct may cause a large increase in the Z-score). The results on the MDT showed that maintenance of the treatment gains mostly occurred for measures that reached proficiency levels of 75% or higher during the treatment phase, such as the correct realization of syllable structures CV and CVC, of place and manner of articulation, and the consistency of productions. This suggests that a child should demonstrate a minimum increase in performance during treatment (in this case a 75% or higher proficiency level) to expect learning (the ability to apply a skill without support; Olswang & Bain, 1994), as reflected by our assessment of maintenance. Additionally, the results on the MDT represent generalization of treatment effects to

untreated items. Maintenance of generalized treatment gains holds great potential for improved intelligibility in daily communication. In the present study, the dose of treatment may have been too low to obtain a 75% proficiency level on all measured features. An extended treatment period or increased intensity may be necessary to obtain the proficiency levels required for broader maintenance of improvement.

Measurements were also chosen to objectify speech motor planning and programming. This was specifically measured ~~in the measures of~~using the MDT, including consistency, fluency and accuracy. For most measures of the MDT there was a strong dip in the scores towards the end of the treatment period. ~~In~~At this time, the boy turned out to be sick. While results on following test administrations did not completely recover, overall, improvement was still apparent.

Measures of speech planning and programming also provide insight ~~in~~into changes in core features of CAS. Consistency ~~is~~was assessed specifically ~~in~~via the repetition of words and non-words, but also in the repetition of syllables ~~in~~on the MDT. Co-articulatory transitions are assessed ~~in~~via the production of clusters ~~and~~. Prosody was to be assessed through the measure of fluency in the MDT. However, this measure was insufficient in this case to objectify prosody. In the definition of CAS as stated by ASHA (2007), the realization of lexical and phrasal stress is named as a marker for inappropriate prosody. As the MDT uses non-word syllables, lexical stress is not assessed. Additionally, in this case, scores for fluency approached ceiling levels very early on, as the boy adopted a strategy where he would produce segmentally simplified, inaccurate sequences in a fluent manner. The structure of the task and the strategy adopted by this boy both contribute to a positive, but incorrect reflection of his prosodic skills in the task results.

The assessment of prosody in the evaluation of SMTA in the treatment of CAS is relevant and should be expanded in future studies. First, assessing all three core features of CAS will provide insight ~~in~~into the effect of treatments on these features. This could lead to better choices in treatment planning, choosing a method that is best for a child at a given point in time. Second, SMTA might be especially effective in the treatment of prosody through the support of music. Speech and music are highly related ~~in~~with respect to prosodic features such as frequency (pitch), duration (rhythm) and intensity (dynamics) (Boutsen, 2003; Hurkmans, 2016; Terband et al., 2019~~) .~~). Because of the similarities between music and speech in prosody and the ~~working~~focus of SMTA ~~at~~on the level of speech planning and programming, it is hypothesized that prosody in children with CAS will improve after

treatment with SMTA. To evaluate this potential effect, specific ~~measure~~measures of prosody at the level of lexical and phrasal stress ~~is~~are needed. Such tasks will be developed to be included in ~~the~~ outcome measurements in future studies.

Conclusion

SMTA, a treatment method that combines speech therapy and music therapy, was introduced ~~in the treatment of CAS. The~~and it's efficacy ~~of this method~~ in the treatment of CAS is now being evaluated in a single subject design study. The present study shows that speech production in the participant improved after treatment, specifically ~~in~~on tasks that relate to CAS and motor speech planning and programming such as consistency and the production of clusters. While intelligibility improved over the study period, objectifying changes in daily communication proved to be difficult. Additional measures may be necessary to gain more insight ~~in~~into treatment effects at the level of communicative participation.

Overall, the results of this first single subject design study provide sufficient support and important ~~leads~~encouragement for further evaluation of SMTA in the treatment of CAS in a proof-of-principle study.

Acknowledgements

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We thank the participant and his parents for their contribution. We thank the speech therapists and music therapists at the rehabilitation centers 'Revalidatie Friesland' and 'Rijndam Revalidatie' for their work in providing treatment and collecting data.

Data access statement

The data can be obtained by contacting the first author.

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Appendix A: Phonetic repertoire

Appendix B: Medical history of the participant

Appendix C: Trained items

Appendix D: Items and scoring of the Modified Diadochokinesis Test

Figures

Figure 1. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and CIred in Picture Naming. PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct, CCVC = Percentage of Initial Clusters Correct, CIred = cluster reduction. T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 2. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and CIred in ~~Nonword~~Non-word Imitation. PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct, CCVC = Percentage of Initial Clusters Correct, CIred = cluster reduction. T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 3. Raw scores and Z-scores for measures of consistency in word and ~~nonword~~non-word repetition. WR = word repetition, NWR = Nonword repetition, T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 4. Raw scores and Z-scores for maximum repetition rate in alternating sequences. T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = Z > + 0.5, clinically relevant change.

Figure 5. Scores for syllable structure on the MDT. Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

Figure 6. Scores for Consistency, Accuracy and Fluency on the MDT. Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

Figure 7. Scores for Place, Manner and Vowel on the MDT and the control task Figure Weights. Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

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Table 1. Structure of a 30 minute treatment session

Phase	Activity	Duration
Introduction	Starting song/ short conversation	3 min
1st exercise	Following protocol in table 2	4 min
2nd exercise	Following protocol in table 2	4 min
Short break	Sing a song or play an instrument	2 min
3rd exercise	Following protocol in table 2	4 min
4th exercise	Following protocol in table 2	4 min
Short break	Sing a song or play an instrument	2 min
5th exercise	Following protocol in table 2	4 min
Conclusion	Make recordings/ sing closing song/ other closing activity	3 min

Note: This structure may vary across children. The time set here per exercise allows for repetition of the entire exercise or practicing a part (word or syllable) of the target item separately before the complete target item.

Table 2. Structure of ~~a~~an SMTA exercise

Step	Participants	Number of repetitions
Introduction of the target word/ sentence	SLP (+ Child)^	1-2 repetitions of target item
Demonstrate target	MT	4-8 repetitions of target item*
Singing	Child + SLP + MT	8-16 repetitions of target item*
Rhythmic chanting	Child + SLP + MT	8-16 repetitions of target item*
Simultaneously speaking	Child + SLP	4-8 repetitions of target item*
Alternately speaking (direct imitation)	Child + SLP	5 repetitions of target item*
Semi-spontaneous speaking (respond to question)	Child + SLP	3 repetitions of target item*

SLP = speech and language pathologist, MT = music therapist. ^At the introduction the child is invited to participate when they are able to repeat the target item correctly. *The number of repetitions is influenced by the length of the target item. A melody usually contains 8 repetitions for words and 4 repetitions for sentences. The melody is sung and chanted at least two times. The numbers here are the minimal number of repetitions that are achieved during one exercise.

Table 3. Schematic representation of outcome measures administered per study phase.

Pre-test	Baseline (5 times)	Weekly testing	Post-test	Follow-up
ICS-Dutch	MDT	MDT	ICS-Dutch	ICS-Dutch
FAN	Figure Weights	Figure Weights	FAN	FAN
CAI			CAI	CAI
CAT			CAT	CAT
			MDT	MDT
			Figure Weights	Figure Weights

ICS-Dutch = Intelligibility in Context Scale – Dutch (McLeod et al., 2012a), FAN = Phonological Analysis for Dutch (Beers, 1995), CAI = Computer Articulation Instrument (Maassen et al., 2019), CAT = Communication Attitude Test (Brutten & Vanryckeghem, 2003), MDT = Modified Diadochokinesis Test (Hurkmans et al., 2012), Figure ~~Weigths~~Weights from the Wechsler Intelligence Scale for Children, ~~vijfde editie; Nederlandstalige bewerking~~Fifth Edition; Dutch Edition (Wechsler, 2018). Full descriptions of these measures are provided under 'Outcome measures' in the ~~method~~methods section.

Table 4. Scores on the ICS-Dutch as scored by the parents of the participant.

ICS-Dutch	T1	T2	T3
Father	3.7	3.4	3.9
Mother	3.6	4	4.1
Parents combined	3.6	3.7	4

T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment).

Table 5. Mean scores of PCCI, PVC and CCVC in spontaneous speech sample

FAN- Dutch	T1	T2	T3
PCCI	85	92	91
PVC	89	97	94
CCVC	40	64	52

PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct, CCVC = Percentage of Initial Clusters Correct. T1 = pre-test, T2 = post-test, T3 = ~~Foflow~~Follow-up (after two months of no treatment).

Table 6. Results for the CAT.

CAT	T1 (Form A)	T2 (Form B)	T3 (Form B)
Raw scores	7	7	3
SD	+ 0,.25	+ 0,.43	- 0.52

SD = standard deviation, T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment).

Note: A higher score indicates a more negative attitude towards their own speech.

Figure 1

Click here to access/download;Figure;SMT in the treatment of CAS - Figure 1 Picture Naming.png

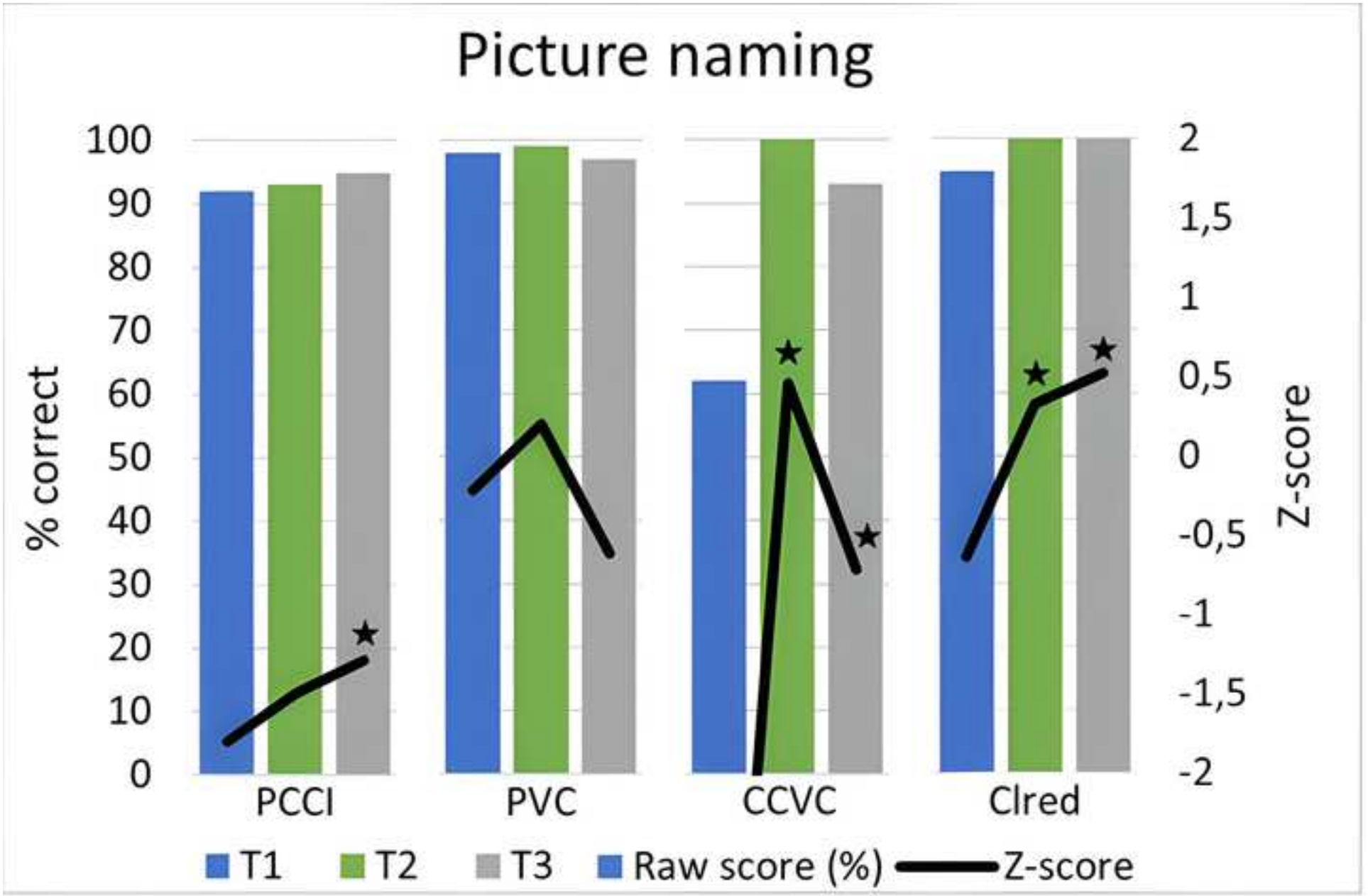


Figure 2

Click here to access/download;Figure;SMT in the treatment of CAS - Figure 2 Non-Word Imitation.png

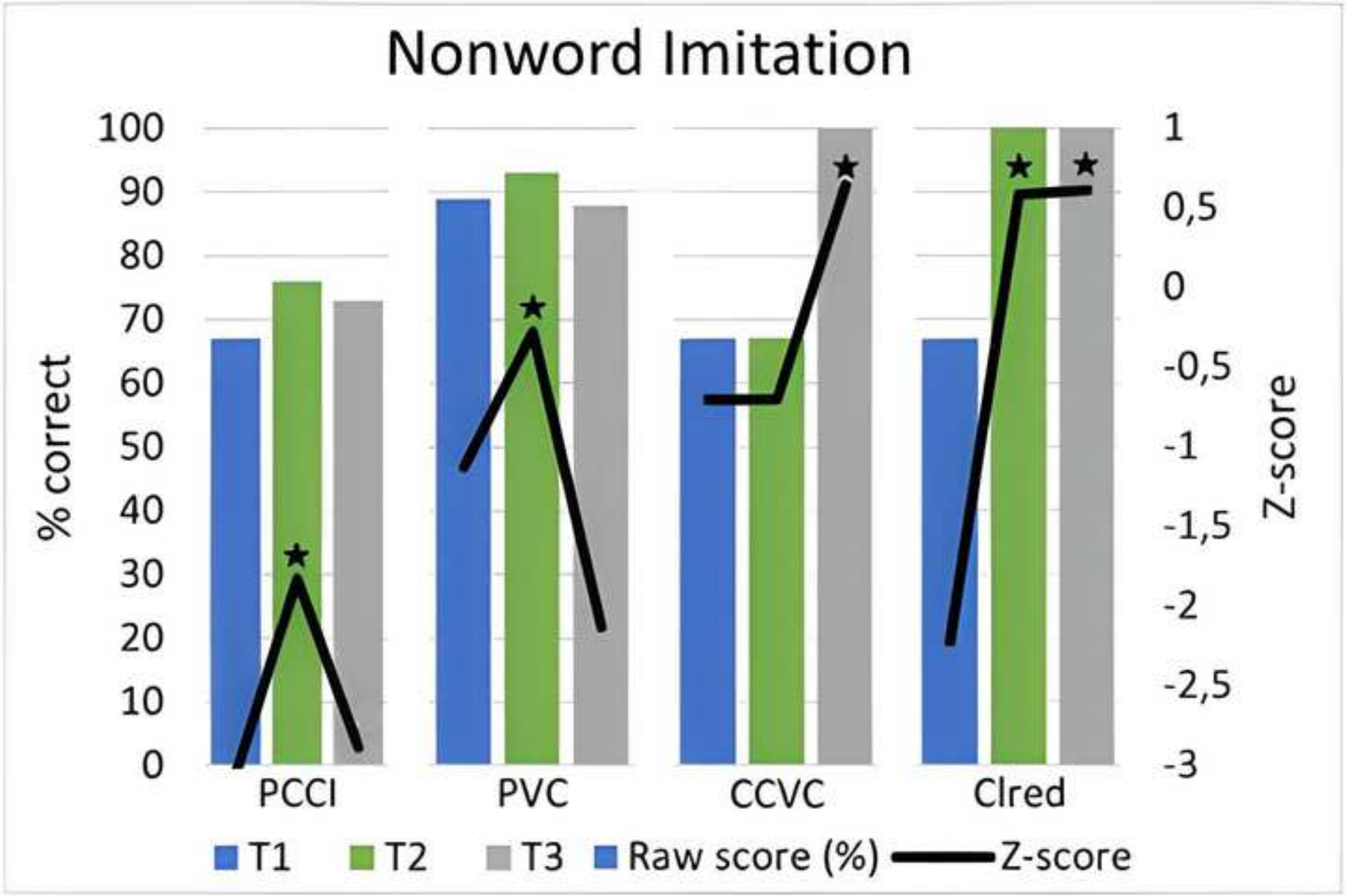


Figure 3

Click here to access/download;Figure;SMT in the treatment of CAS - Figure 3 Consistency.png

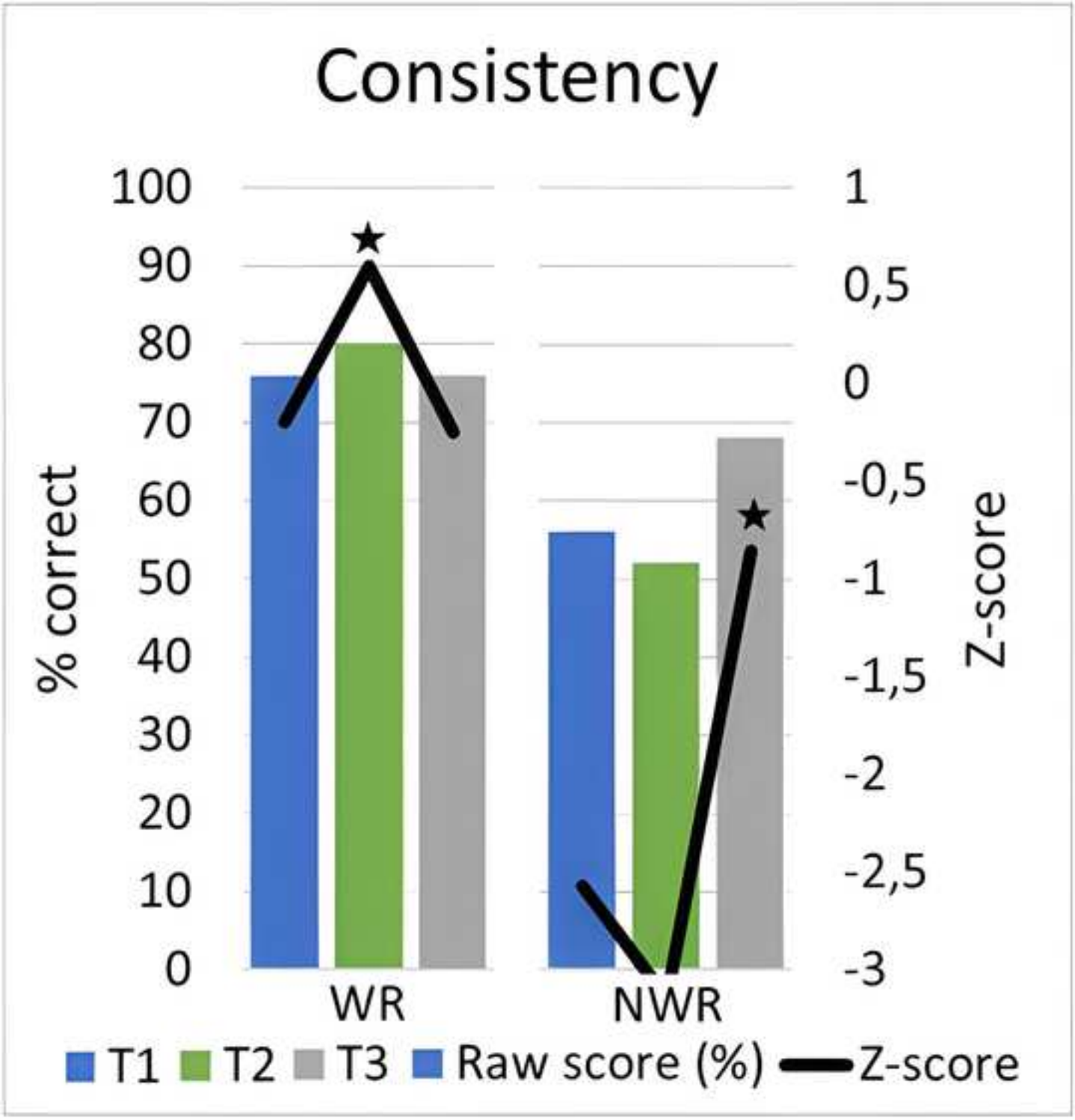


Figure 4

Click here to access/download;Figure;SMT in the treatment of CAS - Figure 4 MRR.png

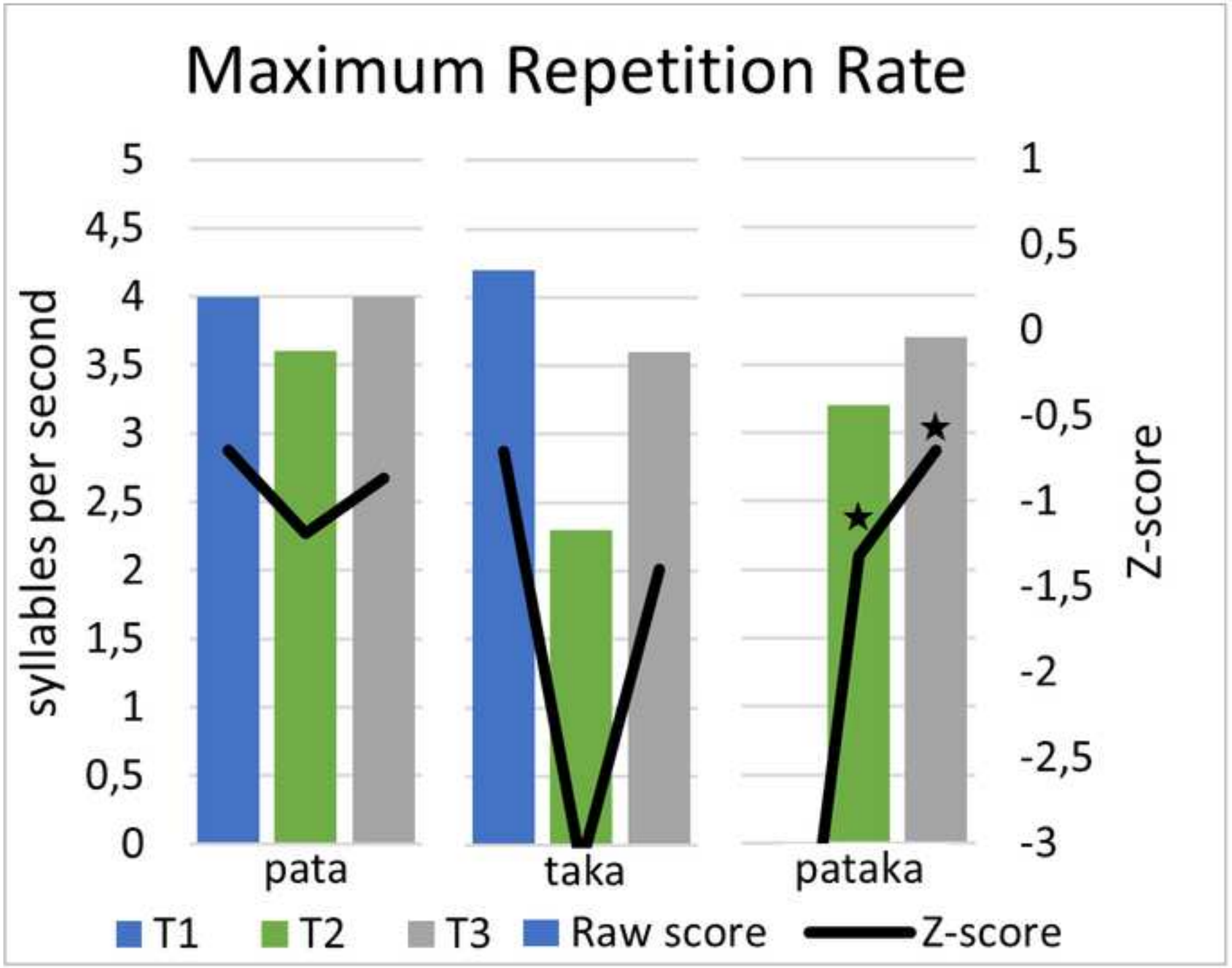


Figure 5

Click here to access/download;Figure;SMT in the treatment of CAS - Figure 5 MDT syll structure.png

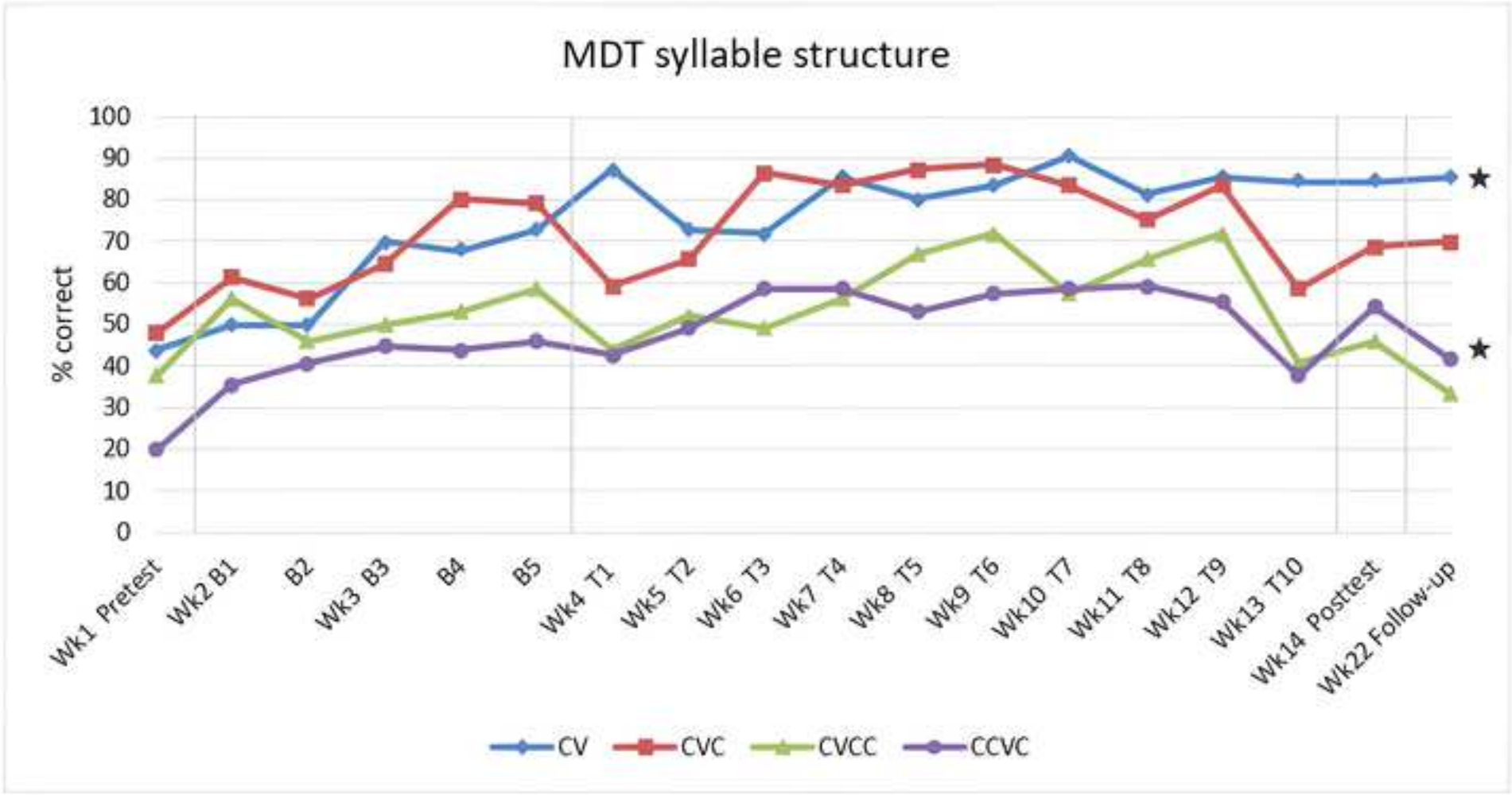


Figure 6

Click here to access/download;Figure;SMT in the treatment of CAS - Figure 6 MDT CAF.png

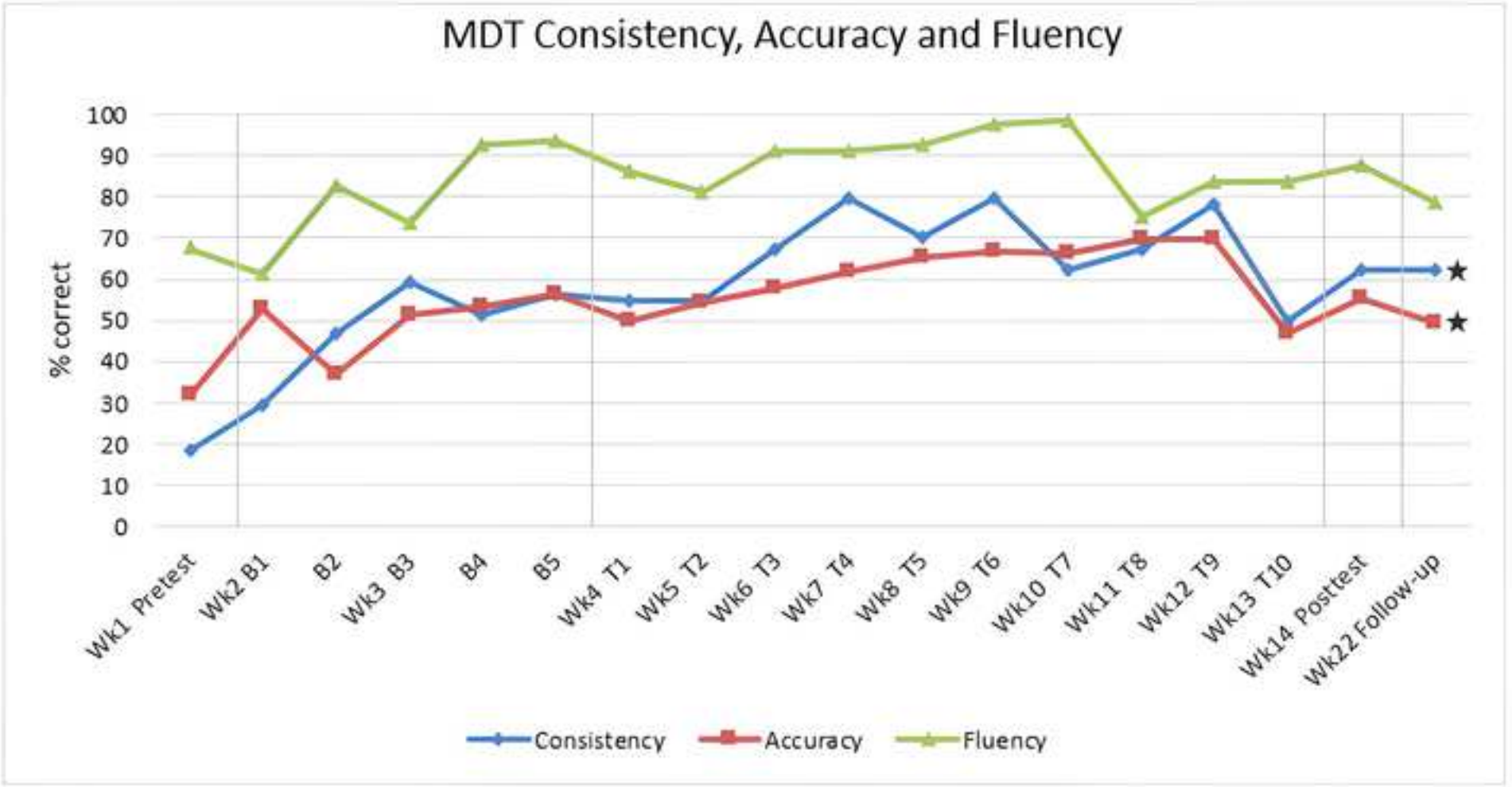
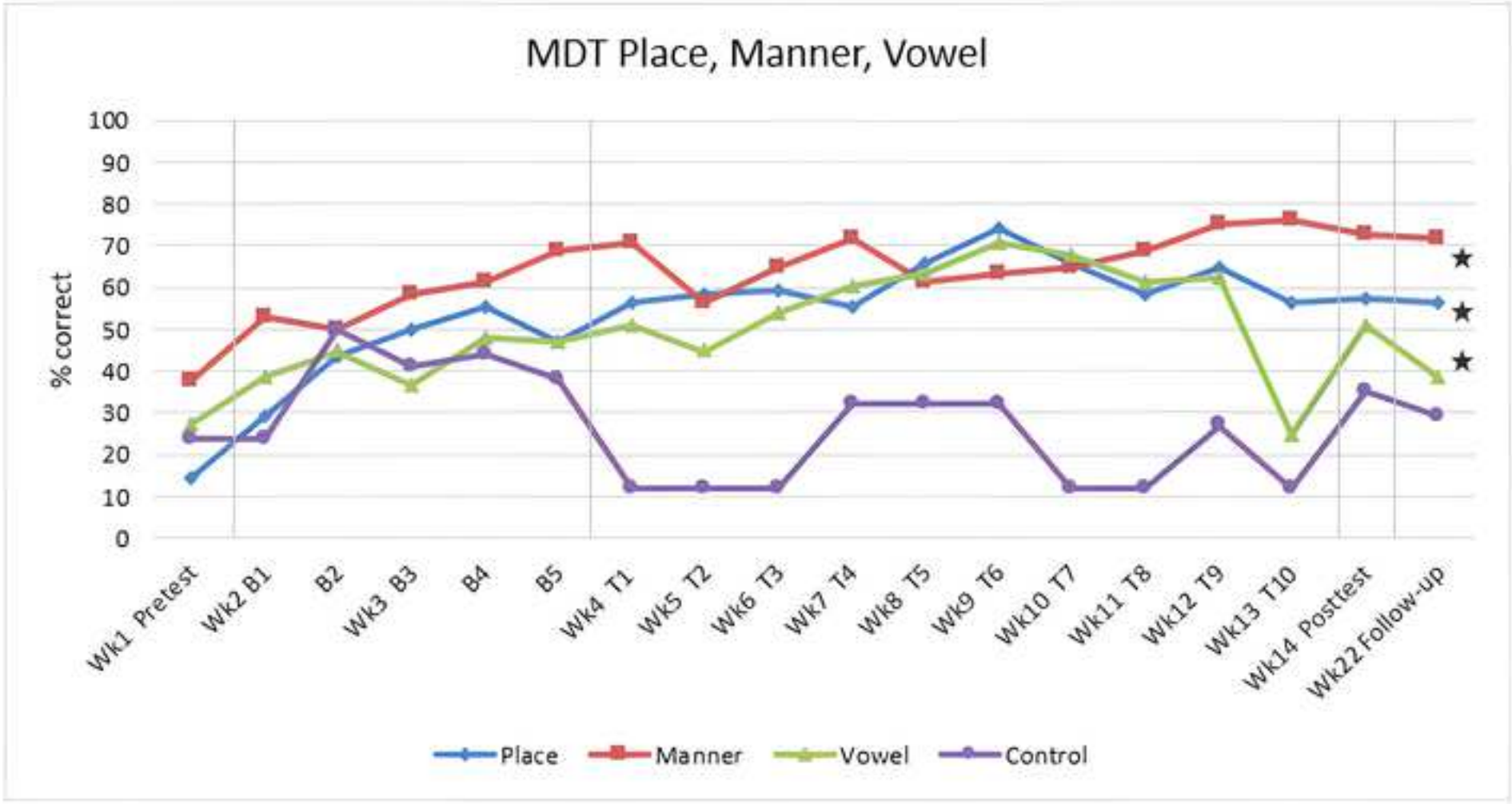


Figure 7

Click here to access/download;Figure;SMT in the treatment of CAS - Figure 7 MDT PMV.png



Reviewers comments	Response
Editor	
Thank you for your considered and significant efforts to reformulate your paper. As you will see below the reviewers are generally positive and have provided significant detail which you can use to finalise your manuscript. Of particular note is changing from the European use a comma to indicate a decimal to the English use of a full stop/ period.	Thank you. We checked the manuscript and changed the commas in the tables into full stops.
Reviewer #1	
This article provides a good example of adapting, modifying, and personalizing a therapy approach designed for the adult population (MIT for Broca's & apraxia) to address the speech sequencing and prosody challenges of children with CAS. The present draft is much improved over the original submission. Removing the ICF as a framework for the article helped the article's flow.	Thank you.
Line 311: It is not clear that the follow-up assessment concluded the no treatment phase. Consider rewording the sentences in 310-311. A suggestion is: A posttest assessment was conducted after the treatment phase and a follow-up assessment was conducted after the two-month no-treatment phase.	We changed this sentence as suggested.
Line 379: The sentence including the stuffed animal needs revision for clarity.	We changed this sentence to reflect that treatment started with an item that was personnally relevant. This in contrast to later weeks were targets were grouped based on speech characteristics.
Discussion or limitations: An interesting observation of the Figures now that the x-axis is clearly labeled and shows the progression over time is that for most measures only the skill that the child produced at the 80-90% proficiency level was maintained. Consider including in the Discussion or Limitation sections that 10 weeks may have been too short or that the child needs to be demonstrating that skill with better proficiency to expect generalization.	Thank you for this observation. We added a paragraph in the limitations section on this subject.
Figure 5: has a random "Grafiekgebied" in a textbox on the figure.	We removed this box from the figure.
Reviewer #2	
The authors have done a thorough job of addressing the reviewers' concerns, overall This study supports future research into the use of SMT with children with CAS	Thank you.

As noted before, this study is well-justified based upon both theories and neurological evidence of how speech and music are perceived, processed, and produced. Practical considerations – engaging children with CAS in the intervention process – are also taken into account. As such, this research is highly appropriate for this journal	
The authors have appropriately addressed the issues related to methods.	Thank you.
The results are clear, overall. However, there is contradictory information about whether or not the child made progress on consonants, in lines 585-593	We changed this section to reflect that there were a fey consonants that showed improvement, thus leading to three categories of consonants in the analysis of spontaneous speech.
In addition, the conclusion about progress in prosody on line 649 is the first mention of progress in this area and does not seem to be well-justified, either here or in the Discussion, where there is more detail about this. I don't feel it's appropriate to claim that the child did make progress in this area.	In this section we use the phrase 'suprasegmental features of speech'. With this phrase we mean to refer to suprasegmental features in a broad sense, not limited to prosody, but including features such as co-articulation, both in the realization of clusters and multisyllabic words. We added a mention of co-articulation to the main text to clarify this. We agree that statements on progress on prosody are not sufficiently supported by our data, as is described in the limitations section.
Figures: Why is there an asterisk for PCCI in Non-Word Imitation at time T3? Line 487 says the progress at T2 was not maintained at follow-up.	Thank you for pointing out this mistake. This asterisk is now deleted from the figure.
Table 2: Change title to "Structure of an SMTA exercise	We changed this as suggested.
Add parenthetical explanation to "Alternately speaking": (direct imitation)	We added this as suggested.
Table 3: Correct spelling of "Weights". If possible, translate the Wechsler citation into English. Change penultimate word to "methods" instead of "method."	We applied these changes as suggested.
Tables 4 & 6: Please use periods in decimals instead of commas, which would be confusing for many readers.	We changed this as suggested.
Table 5: Fix spelling of "Follow-up"	We fixed this.
6. Discussion/Conclusions The discussion has been improved markedly.	Thank you.
Although it is much improved, I still have many suggestions about the writing, listed below by line:	Thank you for your suggestions to improve the writing. We responded to them below.
113-114: Change to: "They also use rate/rhythm control strategies.."	We changed this as suggested.

116: “Additional examples of such..”	In this section we aim to describe the contrast between articulatory-kinematic approaches, such as DTTC and ReST and rate/rhythm control type approaches, such as MIT and SMTA. We have changed this section to clarify this.
119-120: “Both AoS and CAS are described as disorders in the…”	We changed this as suggested.
129: “The results of these studies…”	We changed this as suggested.
142: “…tailored to individual needs”	We changed this as suggested.
152-153: “names of family members, and more formulaic, such as..”	We changed this as suggested.
166: “In that study..”	We changed this as suggested.
170: replace parens around year with [ ]	We changed this as suggested.
172: “To provide the rationale for SMTA in the treatment…”	We changed this as suggested.
174: “..will be discussed:..”	We changed this as suggested.
181: “…showed than an overlap..”	We changed this as suggested.
184-185: “Overlaps between … which is a characteristic of music…” Delete parenthesis before “Boutsen”	We changed this as suggested.
187: “..and intensity are features that combine to express stress in many languages…”	We changed this as suggested.
191: “interventions, which have been summarized..”	We changed this as suggested.
194: “.. insight into” NOTE: Change “insight in” to “insight into” throughout. This is a frequent error.	We fixed this error throughout the manuscript.
195: “..effects of SMTA”	We changed this as suggested.
199: “.. regarded as more..”	We changed this as suggested.
201: “..session, alternating with short musical activities..”	We changed this as suggested.
206-207: “Non-verbal knowledge of performance feedback is …”	We changed this as suggested.
208: “Feedback in the form of knowledge of results..”	We changed this as suggested.
220: “..items are at the word and…”	We changed this as suggested.
222: “..items at the syllable level”	We changed this as suggested.
223: “..targets at the syllable level”	We changed this as suggested.
233: “..as famous or previously used melodies..”	We changed this as suggested.
241: “..Directly thereafter, the child…”	We changed this as suggested.
248: “..introduces turn-taking (direct imitation) with a hand gesture…”	We changed this as suggested.
250: “..used in the introductory phase..”	We changed this as suggested.
262: “..correctly in semi-spontaneous (elicited) speaking…”	We changed this as suggested.
267: “..in the pragmatically intended context…”	We changed this as suggested.
271-274: redundant Basically the same sentence twice. Eliminate one.	We changed this as suggested.
301: “5;8 year-old..”	We changed this as suggested.
302: “..protocol of Iuzzini-Seigel..”	We changed this as suggested.

303-304: "..on a word and non-word repetition task and in spontaneous speech.."	We changed this as suggested.
308: "In the phonological analysis of the child's spontaneous speech.."	We changed this as suggested.
309: "..were produced accurately in less than 50%..."	We changed this as suggested.
310: "..and 100% of the time in initial.."	We changed this as suggested.
312: "..accurately in 33% of occurrences.."	We changed this as suggested.
313: "accurately in between 78%..."	We changed this as suggested.
315-316: "The participant had.."	We changed this as suggested.
318: "..such as difficulties with.."	We changed this as suggested.
331: "..treatment effects."	We changed this as suggested.
337: "showing normal non-verbal psychological development."	We changed this as suggested.
338: "..placed in a specialized.."	We changed this as suggested.
350: "..targets that had been.."	We changed this as suggested.
351: "..items on the sentence level.."	We changed this as suggested.
358: "..was administered.."	We changed this as suggested and changed the sentence overall, following a suggestion from reviewer #1.
359: "..was administered.."	We changed this as suggested.
384: "..consists of the subtests…"	We changed this as suggested.
385: "Word- and Non-Word Repeition.."	We changed this as suggested.
386: "Additionally, the occurrence…"	We changed this as suggested.
387: "cluster reduction (ClRed).."	We changed this as suggested.
395: "The instrument's reliability.."	We changed this as suggested.
397: "The afore-mentioned tasks…"	We changed this as suggested.
401: "consists of items of CV, …"	We changed this as suggested.
403: .. "as accurately as possible"	We changed this as suggested.
409: "..was used in the baseline, weekly testing, and follow-up testing, alongside.."	We changed this as suggested.
417: "..analyze changes on the non-verbal…"	We changed this as suggested.
418: Put info that CAI and CAT are norm-referenced in the Methods, not here. "The CAI and CAT norms were used…"	We changed this as suggested.
419: "Norms from the CAI are divided.."	We changed this as suggested.
420: "..raw scores for the pre-test…"	We changed this as suggested.
431: "The number of (partial) items that were.." Explain what you mean by "partial."	We changed this section to clarify that parts of items were trained seperately at the word level.
432: "..repetitions of items.."	We changed this as suggested.
434-436: Be consistent in use of numbers.	We changed this as suggested.
436: "..personally motivating item of his stuffed animal's name.."	We changed this sentence following suggestions from both reviewers.
439: Delete "the" before items; delete "s" from "sentences"	We changed this as suggested.
440: ".. on multisyllabic words"	We changed this as suggested.
443: ".. treatment. Therefore, this item.."	We changed this as suggested.
448: "..an interdental [n] as a .." (referring to production, not to phoneme)	We changed this as suggested.

449: "..and oral examples … open for the syllable…"	We changed this as suggested.
450: "integrated into the word.."	We changed this as suggested.
451: "..(important); [ŋ] was more easily achieved…	We changed this as suggested.
452: "on. In addition to instances of.."	We changed this as suggested.
457: "Parents reported having had.."	We changed this as suggested.
458: "..received no other speech treatment at all.."	We changed this as suggested.
461: "Combining the scores.."	We changed this as suggested.
467: "..improvement on PCCI.."	We changed this as suggested.
469: ".. realized accurately in 100% of occurrences.."	We changed this as suggested.
472: "..initial clusters, which was obtained.."	We changed this as suggested.
476: "..measures from the CAI showed.."	We changed this as suggested.
483-484: "..a higher score means fewer cluster reductions.."	We changed this as suggested.
485: "On Non-Word Imitation.."	We changed this as suggested.
495: "The DDK task from the CAI, which measures.."	We changed this as suggested.
496-497: "items [pa], [ta], and [ka]."	We changed this as suggested.
499: "..to perform this sequence at pre-test.."	We changed this as suggested.
511: "..was found for the measures.."	We changed this as suggested.
515: "For the scores on the non-verbal.."	We changed this as suggested.
516: "Weights, which is also shown in figure 7, there was no significant.."	We changed this as suggested.
526: "Its potential for the treatment.."	We changed this as suggested.
528: "..frameworks of the neural processing.."	We changed this as suggested.
532: "..this potential, we evaluated the effectiveness.."	We changed this as suggested.
540: "in specific speech-motor tasks."	We changed this as suggested.
541: "word- and non-word repetition, and DDK, with.."	We changed this as suggested.
543: "..progress on measures…"	We changed this as suggested.
544-545: "Improvement on intelligibility and.."	We changed this as suggested.
548: "..in daily life, .."	We changed this as suggested.
553: "..his son's speech.."	We changed this as suggested.
554: "..treatment phases."	We changed this as suggested.
560: "..administrations as well as by the first author.."	We changed this as suggested.
569: "..period, in this case, was…"	We changed this as suggested.
570: "The boy may have had fewer negative experiences.."	We changed this as suggested.
575: "..and attitude towards CAS.."	We changed this sentence to reflect that we mean the attitude towards own speech in children with CAS.
579: "..changes in these features.."	We changed this as suggested.
591: "..and /h/ correctly.."	We changed this as suggested.
593: "..would have resulted in.."	We changed this as suggested.
601: "..in a poor score on cluster reductions, just within normal limits. (Note that scores on cluster reduction were inverted, so that a higher score means less cluster reduction.) After treatment scores.."	We changed this as suggested.

602-604: "..ceiling level. At follow-up, the ceiling level performance on cluster reduction… the boy had now fully.."	We changed this as suggested.
618: "..picture naming versus non-word imitation."	We changed this as suggested.
619: "..on the DDK task,… [pata] and [taka] sequences.."	We changed this as suggested.
622: "..the child was instructed.."	We changed this as suggested.
624-625: "..The production of [pataka], which was impossible for the child at pre-test, did change after treatment."	We changed this as suggested.
628: "In addition to the pre-test, post-test and follow-up, a baseline…"	We changed this as suggested.
629: ".. of the MDT.."	We changed this as suggested.
630: "..sounds on the MDT.."	We changed this as suggested.
631: "Most MDT measured showed a significant.."	We changed this as suggested.
636: Include this sentence ("The improvement in DDK..") in the previous paragraph.	We changed this as suggested.
637: Start new paragraph with "In this study.."	We changed this as suggested.
639: "..some of whom presumably.."	We changed this as suggested.
640: "..that used intoning…"	We changed this as suggested.
641: "..hypothesized about the effect.."	We changed this as suggested.
651: "..the rate/rhythm approach of SMTA."	We changed this as suggested.
652: "..the target group for SMTA.."	We changed this as suggested.
653: "..insight into.."	We changed this as suggested.
657: "case study of a series within a proof-of-principle study.."	We changed this as suggested.
659: "..evidence by itself."	We changed this as suggested.
662: "..sufficient encouragement for follow-up studies."	We changed this as suggested.
663-664: "..reflect intelligibility in daily.."	We changed this as suggested.
665-666: "In addition to concerns about the scoring of the ICS-Dutch.."	We changed this as suggested.
668-670: "provides insight into intelligibility.. participation has changed."	We changed this as suggested.
671: "..insight into.."	We changed this as suggested.
672: "..Focus on the Outcomes.."	We changed this as suggested.
673: "..insight into changes.."	We changed this as suggested.
677-678: "..at the age of… reflecting nearly completed.."	We changed this as suggested.
679-680: "..one error affecting the raw score may cause a large drop in the Z-score…. increase in the Z-score).	We changed this as suggested.
682-683: "measured using the MDT, including.."	We changed this as suggested.
685: "At this time, the boy.."	We changed this as suggested.
689-690: "..insight into changes… Consistency was assessed specifically via the repetition…"	We changed this as suggested.
691-693: "..syllables on the MDT…. are assessed via the production of clusters. Prosody was to be assessed via the fluency measure on the MDT."	We changed this as suggested.
695: "..the realization of lexical.."	We changed this as suggested.

702: “..insight into..”	We changed this as suggested.
706: “..related with respect to prosodic..”	We changed this as suggested.
708: “..the focus on the SMTA on the level..”	We changed this as suggested.
710: “..measures of prosody..”	We changed this as suggested.
711: “..lexical and phrasal stress are needed.”	We changed this as suggested.
712: “included in outcome measurements..”	We changed this as suggested.
715-716: “.. was introduced to evaluated the efficacy of …CAS in a single subject design study.”	We changed the start of the conclusion and combined the first two sentences, to reflect that this manuscript is intended to introduce SMTA as a method in the treatment of CAS and that it's efficacy is now being evaluated in a single subject design study.
718: “..specifically on tasks..”	We changed this as suggested.
722: “..insight into treatment..”	We changed this as suggested.
724-725: “..important encouragement for further..”	We changed this as suggested.

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