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

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Abstract:	<p>Purpose</p> <p>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.</p> <p>Method</p> <p>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.</p> <p>Results</p> <p>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.</p> <p>Conclusion</p> <p>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.</p>
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1 **Speech and music therapy in the treatment of CAS: An introduction and a case study**

2

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29 Mirjam van Tellingen: Conceptualization, data curation, formal analysis, investigation, methodology,
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38

39 **Conflict of interest statement**

40 The authors report no conflict(s) of interest.

41 **Abstract**

42 Purpose

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

46 Method

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

52 Results

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

56 Conclusion

57 This case study showed that speech production of the participant improved after treatment with SMTA.
58 Although intelligibility as measured with the ICS-Dutch improved over the study period, objectifying
59 changes at the level of intelligibility in daily communication proved to be difficult. Additional measures
60 may be necessary to gain more insight into treatment effects at this level.
61 Overall, the results of this first case study provide sufficient support and important leads for further
62 evaluation of SMTA in the treatment of CAS in a proof-of-principle study.

63

64 **Introduction**

65 Childhood Apraxia of Speech (CAS) is a speech sound disorder classified as a subtype of motor
66 speech disorder (Shriberg et al., 2010). In CAS, a core impairment at the level of speech motor
67 planning and programming results in errors in the production of speech sounds and prosody.

68 Inappropriate prosody, inconsistency and disrupted coarticulation are widely described as three key
69 features of CAS (e.g., American Speech-Language-Hearing Association, 2007; Shriberg et al.,

70 1997abc; Terband et al., 2019). Inappropriate prosody, consonant and vowel errors, and voicing and

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

74 Treatments for CAS address one or more of the three core features of CAS. In Dynamic Temporal and
75 Tactile Cueing (DTTC; Strand, 2020; Strand et al., 2006), a method for children with more severe
76 CAS, all three features are targeted through a focus on movements rather than phonemes, varying
77 prosody and high numbers of repetitions. Rapid Syllable Transition treatment (ReST; Ballard et al.,
78 2010), which is used in older children with less severe CAS, addresses all three features through the
79 focus on sounds, beats and smoothness. Studies report positive outcomes on ratings of production
80 accuracy for DTTC (e.g., Maas et al., 2012; Maas & Farinella, 2012; Strand et al., 2006) and on
81 segmental accuracy and lexical stress for ReST (Ballard et al., 2012; McCabe et al., 2014). While
82 results of these studies for different features of CAS are promising, McCabe and colleagues (2014)
83 showed that the children improved on either lexical stress production or segmental accuracy, but they
84 remained unable to simultaneously produce both correct stress patterns and correct segments. This is
85 known to be a major challenge in developmental speech disorders (Howard, 2007).

86 Both DTTC and ReST can be described as articulatory-kinematic approaches, using interventions
87 such as visual and tactile cues as well as feedback on the knowledge of performance. These methods
88 also use some rate/rhythm control strategies, such as reduced speech rate and specific drill of lexical
89 stress. Other methods, such as Melodic Intonation Therapy (MIT; Albert et al., 1973; Helfrich-Miller,
90 1984) and Speech-Music Therapy for Aphasia (SMTA; De Bruijn et al., 2005; Hurkmans et al., 2018),
91 are primarily described as rate/rhythm control type approaches. The interventions in these methods
92 are aimed at speech rate, stress, and intonation. MIT and SMTA were originally developed for adults
93 with non-fluent aphasia and Apraxia of Speech (AoS; Hurkmans et al., 2015; Merrett et al., 2014).

94 Both AoS and CAS are described as disorders in the planning and programming of speech
95 movements (American Speech-Language-Hearing Association, 2007; Hurkmans, 2016) and share
96 various characteristics, such as inconsistent errors in the realization of phonemes, syllable
97 segregation, vowel distortions, groping and effect of articulatory complexity (Iuzzini-Seigel & Murray,
98 2017; Ziegler, 2008). Therefore, rate/rhythm control approaches might be effective in the treatment of
99 CAS.

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

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

109

110 SMTA

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

118 There are two lines of treatment: a speech therapy line and a music therapy line, that are conducted
119 simultaneously. Speech therapy includes three levels: (1) syllables, (2) words and (3) sentences.

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

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

141 SMTA is originally based on various similarities between language and music, such as shared
142 hierarchical structures (Hurkmans, 2016; Patel, 2003; Peretz & Zatorre, 2003) and shared neural
143 processing (e.g. Brown et al., 2006; see Hurkmans [2016] for further discussion). SMTA has been
144 shown to improve speech production at the level of motor planning and programming (Hurkmans,
145 2015). To provide the rationale for SMTA in the treatment of CAS, three theoretical frameworks on
146 music and speech and the potential working mechanisms of music in the treatment of speech will be
147 discussed: (1) similarities between and overlap in the processing of speech and music, (2) overlap in
148 prosodic features in music and speech, and (3) mechanisms of music with regards to motivation and
149 mood.

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

160 interventions, which have been summarized by Merrett, Peretz and Wilson (2014) as one of the
161 possible working mechanisms of MIT. In short, music is believed to have a positive effect on mood and
162 motivation, which may contribute to the effect of interventions that utilize musical elements. Together,
163 these frameworks provide insight into the potential mechanisms that contribute to the effects of SMTA.
164 SMTA encompasses all principles of motor learning that are recommended in the treatment of CAS
165 (Maas et al., 2014; Murray et al., 2014; Strand, 2020). The use of music in SMTA allows for a high
166 number of trials per session, which is crucial for motor learning (Maas et al., 2014; Strand, 2020), as
167 singing (including a minimum of 20 trials per exercise) is regarded as more pleasant than realizing a
168 high number of trials in a drill-type exercise. Usually up to five different exercises are conducted during
169 a treatment session, alternating with small musical activities, such as singing a song or playing an
170 instrument. Children are given autonomy within the session as they are invited to select the targets
171 they want to practice and choose the musical activities they wish to engage in. This autonomy and the
172 highly relevant target items can both contribute to increased motivation (Strand, 2020; Wulf et al.,
173 2018). During an exercise, verbal feedback is kept to a minimum, to avoid disruptions of the flow of the
174 exercise. Non-verbal knowledge of performance feedback is provided by focusing the child's attention
175 on the provided oral example in which the speech therapist may emphasize a specific movement.
176 Feedback in the form of knowledge of results is also provided non-verbally and focusses on accurate
177 realizations of the target word. Placing the child's attention on accurate realizations raises their
178 expectancies of their own ability. Before and at the end of an exercise, the functional use of the target
179 is emphasized, to direct the attention to an external focus on results. This focus on feedback at the
180 level of knowledge of results is recommended in the treatment of children with CAS to stimulate
181 learning and retention (Strand, 2020) and has consistently been shown to enhance learning
182 regardless of task, age, skill level, or (dis)ability (Wulf et al., 2018).

183

184 Treatment protocol

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

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

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

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

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

225

226 *Insert table 1 and 2 around here.*

227

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

235

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

243

244 **Case study**

245 Method

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

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

259

260 Participant

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

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

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

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

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

299

300 Intervention

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

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

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

313

314 Outcome measures

315 A schematic overview of the outcome measures and timing of administration is presented in table 3.

316 The primary outcome measure was chosen to reflect intelligibility in daily communication, in line with
317 the core objective of speech therapy, to support the child in communicative participation in society

318 (Hustad, 2012). The selected measure was the Intelligibility in Context Scale – Dutch (ICS-Dutch;

319 McLeod et al., 2012a). In this questionnaire, parents rate the intelligibility of their child in contact with

320 various communication partners, such as family members, peers, teachers, and strangers on a five-

321 point scale. Reliability and validity for this instrument was assessed for the original English version

322 (McLeod et al., 2012b) and the Dutch version used in the current study (McLeod, 2020; Van Doornik et

323 al., 2018) and found to be adequate. Both parents filled out the ICS-Dutch independent of one

324 another.

325 Further outcome measures were selected to reflect speech motor abilities in various tasks, such as

326 spontaneous speech, picture naming, non-word repetition and DDK.

327 A speech sample was collected and analyzed using the Phonological Analyses for Dutch

328 (Fonologische Analyse voor het Nederlands (FAN); Beers, 1995; Beers & Masereeuw, 2022). The

329 sample was elicited through a series of standardized questions on topics such as school, hobbies, and

330 vacation. The first 100 unique words in the sample were transcribed and analyzed. In this task

331 Percentage Consonants Correct in syllable initial position (PCCI) and Percentage Vowels Correct

332 (PVC) were calculated, as well as percentage of clusters correct in syllable initial position (CCVC).

333 The Computer Articulation Instrument (CAI; Maassen et al., 2019) was used to assess speech in

334 specific tasks. The CAI consists of the subtests Picture Naming, Non-word Imitation, Word- and Non-

335 Word Repetition and DDK. For Picture Naming and Non-word Imitation PCCI, PVC and CCVC were

336 calculated. Additionally, the occurrence of cluster reduction (Clred) was calculated. For Word- and

337 non-wordrepetition consistency was calculated and for DDK the measure was Maximum Repetition

338 Rate (MRR). The CAI is norm-referenced and has been shown to have sufficient to good reliability and

339 validity for the assessment of speech development in Dutch children ages 2-7 (van Haaften et al.,
340 2019). The CAI norms are divided into age groups that span four months for the younger groups and
341 six months for the two oldest groups. For this study, raw scores for the pre-test and post-test were
342 compared with the norms for children aged 5;8-5;11 and raw scores for follow-up were compared to
343 the norms for children aged 6;0-6;5 (van Haaften et al., 2019).

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

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

360 A non-verbal control task was used to control for developmental progress. This was the task Figure
361 Weights from the Wechsler Intelligence Scale for Children, Fifth Dutch edition (WISC-V-NL; Wechsler,
362 2018). The Figure Weights task was used in the baseline, weekly testing, and follow-up testing,
363 alongside the MDT. Reliability and validity of the WISC-V-NL are adequate (Wechsler, 2018).

364

365

Insert table 3 around here

366

367 Analysis

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

372

373 Results

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

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

405
406 Results for intelligibility as measured with the ICS-Dutch, are presented in table 4. Combining the
407 scores of both parents, the results suggested some improvement over the study period. Scores of the
408 father and mother individually show different patterns.

409

410 *Insert table 4 around here.*

411
412 The analysis of a sample of spontaneous speech with the FAN suggested improvement on PCCI over
413 the treatment period, as presented in table 5. Improvement was found for /x/ and /s/, which were
414 realized accurate in 100% of occurrences after treatment, which was maintained at follow-up. The
415 realization of /d/ improved after treatment and improved further over the follow-up period. For the
416 consonant /r/ there was some improvement after treatment, which was not maintained at follow-up.
417 Improvement on PVC and the production of initial clusters, which was obtained after treatment, was
418 not maintained at follow-up.

419

420 *Insert table 5 around here.*

421 Tasks and measures from the CAI showed varying results. The CAI does not provide critical
422 differences, therefore a change of $Z > + 0.5$ was set as the norm for clinically relevant change. Z-
423 scores were calculated using the means and SDs published by Van Haaften et al. (2019). In Picture
424 Naming, presented in figure 1, there was clinically relevant change for PCCI over the study period ($Z +$
425 0.51 at follow-up compared to pre-test), but no clinically relevant changes for PVC ($Z + 0.42$ over
426 treatment period). The production of initial clusters in Picture Naming improved after treatment ($Z +$
427 5.46), and the gains were mostly maintained at follow-up ($Z + 4.29$). Results for cluster reductions in
428 the same task, for which scores were inverted to reflect that a higher score means fewer cluster

429 reductions, showed clinically relevant change over the treatment period, reaching ceiling level after
430 treatment ($Z + 0.97$), which was maintained at follow-up ($Z + 1.16$). On Non-Word Imitation, presented
431 in figure 2, results for PCCI showed clinically relevant change after treatment ($Z + 1.22$), but this was
432 not maintained at follow-up ($Z + 0.16$). Results for PVC showed an increase from below average to
433 within normal limits directly after treatment ($Z + 0.85$), but this improvement was not maintained at
434 follow-up ($Z - 1.0$). The production of initial clusters in Non-Word Imitation did not change directly after
435 therapy, but did improve at follow-up ($Z + 1.34$). Results for cluster reduction in this task showed
436 clinically relevant change, reaching ceiling level after treatment ($Z + 2.8$), which was maintained at
437 follow-up ($Z + 2.83$). Results for consistency are presented in figure 3. Word consistency increased
438 directly after treatment ($Z + 0.80$), but this improvement was not maintained at follow-up. Non-word
439 consistency did not increase directly after treatment, but improvement was apparent at follow-up ($Z +$
440 1.71). The DDK task from the CAI, which measures maximum repetition rate (MRR; syllables per
441 second) showed no change for the sequential items /pa/, /ta/ and /ka/. Results for the alternating items
442 are presented in figure 4. For the items /pata/ and /taka/, results were lower directly after treatment (Z
443 $- 0.48$ and $Z - 2.42$) and at follow-up ($Z - 0.16$ and $Z - 0.69$). For /pataka/ the scores improved after
444 treatment ($Z + 3.59$) from not being able to perform this sequence at pre-test to within normal limits at
445 follow-up ($Z + 4.21$).

446 *Insert figures 1,2, 3 and 4 around here*

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

450 *Insert table 6 around here*

451
452 Results on several measures of the MDT showed change after treatment and over the study period.
453 For syllable structure, there was a significant gradual improvement on CV (Kendall $\tau = 0.633$, $p < .01$)
454 and CCVC (Kendall $\tau = 0.396$, $p < .05$) structures as shown in figure 5. Results for the structures CVC
455 (Kendall $\tau = 0.290$, $p > .05$) and CVCC (Kendall $\tau = 0.132$, $p > .05$) showed no significant change over
456 the study period. Significant gradual improvement was found for the measures of accuracy (Kendall τ

457 = 0.433, $p < .05$) and consistency (Kendall $\tau = 0.447$, $p < .05$), but not for fluency (Kendall $\tau = 0.211$, p
458 $> .05$) as shown in figure 6. Figure 7 shows significant gradual improvement for the measures place
459 (Kendall $\tau = 0.513$, $p < .01$), manner (Kendall $\tau = 0.656$, $p < .01$) and vowel (Kendall $\tau = 0.356$, $p <$
460 $.05$). For the scores on the non-verbal control task Figure Weights, which is also shown in figure 7,
461 there was no significant improvement over the study period (Kendall $\tau = -0.210$, $p > .05$).

462

463 *Insert figure 5, 6 and 7 around here*

464

465 **Discussion**

466

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

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

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

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

512 Intelligibility and attitude towards own speech are known to be impacted by inappropriate prosody,
513 consonant and vowel, errors and voicing and nasality errors (Chenausky et al., 2022; Klopfenstein,
514 2009; McCabe et al., 2014; Hustad, 2012). This suggests that the improvement in intelligibility and
515 attitude in this study may be caused by changes in this type of features related to speech motor

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

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

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

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

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

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

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

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

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

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

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

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

589

590 Limitations and directions for future research

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

597 The measurements in this study were chosen to reflect intelligibility in daily communication, as well as
598 speech motor planning and programming. Objectifying change in intelligibility in daily communication
599 proved to be difficult. In addition to concerns about the scoring of the ICS-Dutch by the father in this
600 specific case, this instrument is vulnerable to bias, as parents are aware of the timing of treatment.

601 Additionally, this instrument provides insight into intelligibility in daily communication, but does not
602 measure whether communicative participation has changed. Changes in communicative participation
603 could be expected when both intelligibility and attitudes towards speech improve, but the current
604 measures did not provide insight into participation. An additional measure, such as Focus on the

605 Outcomes of Communication Under Six (FOCUS; Thomas- Stonell et al., 2013) might be useful in
606 future studies to get a broader insight into changes in communicative participation after treatment.
607 Results on the CAI show that for some measures, a small change in the raw score may result in a far
608 greater change in Z-score. This is caused by ceiling effects on these measures. Typically developing
609 children at the age of the participant score close to 100% on most measures, reflecting nearly
610 completed motor-speech development (although refinement of these skills continues for longer;
611 Ballard et al., 2012). Therefore, one error affecting the raw score, may cause a large drop in the Z-
612 score for such a measure (or one more item correct may cause a large increase in the Z-score).
613 The results on the MDT showed that maintenance of the treatment gains mostly occurred for
614 measures that reached proficiency levels of 75% or higher during the treatment phase, such as the
615 correct realization of syllable structures CV and CVC, of place and manner of articulation, and the
616 consistency of productions. This suggests that a child should demonstrate a minimum increase in
617 performance during treatment (in this case a 75% or higher proficiency level) to expect learning (the
618 ability to apply a skill without support; Olswang & Bain, 1994), as reflected by our assessment of
619 maintenance. Additionally, the results on the MDT represent generalization of treatment effects to
620 untreated items. Maintenance of generalized treatment gains holds great potential for improved
621 intelligibility in daily communication. In the present study, the dose of treatment may have been too low
622 to obtain a 75% proficiency level on all measured features. An extended treatment period or increased
623 intensity may be necessary to obtain the proficiency levels required for broader maintenance of
624 improvement.

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

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

635 (2007), the realization of lexical and phrasal stress is named as a marker for inappropriate prosody. As
636 the MDT uses non-word syllables, lexical stress is not assessed. Additionally, in this case, scores for
637 fluency approached ceiling levels very early on, as the boy adopted a strategy where he would
638 produce segmentally simplified, inaccurate sequences in a fluent manner. The structure of the task
639 and the strategy adopted by this boy both contribute to a positive, but incorrect reflection of his
640 prosodic skills in the task results.

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

653

654 **Conclusion**

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

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

664

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670

671 **Data access statement**

672 The data can be obtained by contacting the first author.

673

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832

833 **Appendix A: Phonetic repertoire**

834

835 **Appendix B: Medical history of the participant**

836

837 **Appendix C: Trained items**

838

839 **Appendix D: Items and scoring of the Modified Diadochokinesis Test**

840

841 **Figures**

842 **Figure 1. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and Clred in Picture**

843 **Naming.** PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels
844 Correct, CCVC = Percentage of Initial Clusters Correct, Clred = cluster reduction. T1 = pre-test, T2 =
845 post-test, T3 = follow-up (after two months of no treatment), ★ = $Z > + 0.5$, clinically relevant change.

846

847

848 **Figure 2. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and Clred in Non-word**

849 **Imitation.** PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels
850 Correct, CCVC = Percentage of Initial Clusters Correct, Clred = cluster reduction. T1 = pre-test, T2 =
851 post-test, T3 = follow-up (after two months of no treatment), ★ = $Z > + 0.5$, clinically relevant change.

852

853 **Figure 3. Raw scores and Z-scores for measures of consistency in word and non-word**

854 **repetition.** WR = word repetition, NWR = Nonword repetition, T1 = pre-test, T2 = post-test, T3 =
855 follow-up (after two months of no treatment), ★ = $Z > + 0.5$, clinically relevant change.

856

857 **Figure 4. Raw scores and Z-scores for maximum repetition rate in alternating sequences.** T1 =

858 pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = $Z > + 0.5$, clinically
859 relevant change.

860

861 **Figure 5. Scores for syllable structure on the MDT.** Wk = week, B = baseline, T = treatment, ★ =

862 significant $p < .05$, Kendall's tau test.

863

864 **Figure 6. Scores for Consistency, Accuracy and Fluency on the MDT.** Wk = week, B = baseline, T

865 = treatment, ★ = significant $p < .05$, Kendall's tau test.

866

867 **Figure 7. Scores for Place, Manner and Vowel on the MDT and the control task Figure Weights.**

868 Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

869

870

871 **Table 1. Structure of a 30 minute treatment session**
 872

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

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

876 **Table 2. Structure of an SMTA exercise**
 877

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*

878 SLP = speech and language pathologist, MT = music therapist. ^At the introduction the child is invited
 879 to participate when they are able to repeat the target item correctly. *The number of repetitions is
 880 influenced by the length of the target item. A melody usually contains 8 repetitions for words and 4
 881 repetitions for sentences. The melody is sung and chanted at least two times. The numbers here are
 882 the minimal number of repetitions that are achieved during one exercise.
 883

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

895 **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

896 PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct,
897 CCVC = Percentage of Initial Clusters Correct. T1 = pre-test, T2 = post-test, T3 = Follow-up (after two
898 months of no treatment).
899

900 **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

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

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

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

2

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39 **Conflict of interest statement**

40 The authors report no conflict(s) of interest.

41 **Abstract**

42 Purpose

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

46 Method

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

52 Results

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

56 Conclusion

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

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

63

64 **Introduction**

65 Childhood Apraxia of Speech (CAS) is a speech sound disorder classified as a subtype of motor
66 speech disorder (Shriberg et al., 2010). In CAS, a core impairment at the level of speech motor
67 planning and programming results in errors in the production of speech sounds and prosody.

68 Inappropriate prosody, inconsistency and disrupted coarticulation are widely described as three key
69 features of CAS (e.g., American Speech-Language-Hearing Association, 2007; Shriberg et al.,

70 1997abc; Terband et al., 2019). Inappropriate prosody, consonant and vowel errors, and voicing and

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

74 Treatments for CAS address one or more of the three core features of CAS. In Dynamic Temporal and
75 Tactile Cueing (DTTC; Strand, 2020; Strand et al., 2006), a method for children with more severe
76 CAS, all three features are targeted through a focus on movements rather than phonemes, varying
77 prosody and high numbers of repetitions. Rapid Syllable Transition treatment (ReST; Ballard et al.,
78 2010), which is used in older children with less severe CAS, addresses all three features through the
79 focus on sounds, beats and smoothness. Studies report positive outcomes on ratings of production
80 accuracy for DTTC (e.g., Maas et al., 2012; Maas & Farinella, 2012; Strand et al., 2006) and on
81 segmental accuracy and lexical stress for ReST (Ballard et al., 2012; McCabe et al., 2014). While
82 results of these studies for different features of CAS are promising, McCabe and colleagues (2014)
83 showed that the children improved on either lexical stress production or segmental accuracy, but they
84 remained unable to simultaneously produce both correct stress patterns and correct segments. This is
85 known to be a major challenge in developmental speech disorders (Howard, 2007).

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

100 2017; Ziegler, 2008). Therefore, rate/rhythm control approaches might be effective in the treatment of
101 CAS.

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

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

111

112 SMTA

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

120 There are two lines of treatment: a speech therapy line and a music therapy line, that are conducted
121 simultaneously. Speech therapy includes three levels: (1) syllables, (2) words and (3) sentences.

122 These levels allow for a focus on movements, rather than individual speech sounds. As an exception,
123 vowels may be practiced in isolation at the first level. Target items on the word and sentence levels
124 are designed both to fit the speech targets based on speech assessment, as well as to be functionally
125 relevant (and therefore motivating) for the individual child in daily communication. For example, when
126 a child has difficulty producing consonant clusters, and their brother's name is 'Steven', this could
127 make this name an excellent target item. Items may be both personal, such as names of family
128 members, and more formulaic ~~items~~, such as 'thank you'. Music therapy follows a structured
129 procedure that starts with singing, followed by rhythmical chanting and speaking, which is derived from

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

143 SMTA is originally based on various similarities between language and music, such as shared
144 hierarchical structures (Hurkmans, 2016; Patel, 2003; Peretz & Zatorre, 2003) and shared neural
145 processing (e.g. Brown et al., 2006; see Hurkmans [\(2016\)](#) for further discussion). SMTA has been
146 shown to improve speech production at the level of motor planning and programming (Hurkmans,
147 2015). To provide the rationale [effor](#) SMTA in the treatment of CAS, three theoretical frameworks on
148 music and speech and the potential working mechanisms of music in the treatment of speech will be
149 discussed: (1) similarities between and overlap in the processing of speech and music, (2) overlap in
150 prosodic features in music and speech, and (3) mechanisms of music with regards to motivation and
151 mood.

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

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

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

186

187 Treatment protocol

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

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

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

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

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

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

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

229

230 *Insert table 1 and 2 around here.*

231

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

239

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

248

249 **Case study**

250 Method

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

264

265 Participant

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

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

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

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

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

304

305 Intervention

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

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

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

319

320 Outcome measures

321 A schematic overview of the outcome measures and timing of administration is presented in table 3.

322 The primary outcome measure was chosen to reflect intelligibility in daily communication, in line with

323 the core objective of speech therapy, to support the child in communicative participation in society

324 (Hustad, 2012). The selected measure was the Intelligibility in Context Scale – Dutch (ICS-Dutch;

325 McLeod et al., 2012a). In this questionnaire, parents rate the intelligibility of their child in contact with

326 various communication partners, such as family members, peers, teachers, and strangers on a five-

327 point scale. Reliability and validity for this instrument was assessed for the original English version

328 (McLeod et al., 2012b) and the Dutch version used in the current study (McLeod, 2020; Van Doornik et

329 al., 2018) and found to be adequate. Both parents filled out the ICS-Dutch independent of one

330 another.

331 Further outcome measures were selected to reflect speech motor abilities in various tasks, such as

332 spontaneous speech, picture naming, non-word repetition and DDK.

333 A speech sample was collected and analyzed using the Phonological Analyses for Dutch

334 (Fonologische Analyse voor het Nederlands (FAN); Beers, 1995; Beers & Masereeuw, 2022). The

335 sample was elicited through a series of standardized questions on topics such as school, hobbies, and

336 vacation. The first 100 unique words in the sample were transcribed and analyzed. In this task

337 Percentage Consonants Correct in syllable initial position (PCCI) and Percentage Vowels Correct

338 (PVC) were calculated, as well as percentage of clusters correct in syllable initial position (CCVC).

339 The Computer Articulation Instrument (CAI; Maassen et al., 2019) was used to assess speech in
340 specific tasks. The CAI consists of the subtests Picture Naming, ~~Nonword~~Non-word Imitation, Word-
341 and ~~nonwordrepetition~~Non-Word Repetition and DDK. For Picture Naming and Non-word Imitation
342 PCCI, PVC and CCVC were calculated. Additionally, the occurrence of ~~Clusterreduction~~cluster
343 reduction (Clred) was calculated. For Word- and ~~nonwordrepetition~~non-wordrepetition consistency was
344 calculated and for DDK the measure was Maximum Repetition Rate (MRR). The CAI has norm-
345 referenced and has been shown to have sufficient to good reliability and validity for the assessment of
346 speech development in Dutch children ages 2-7 (van Haaften et al., 2019). The CAI norms are divided
347 into age groups that span four months for the younger groups and six months for the two oldest
348 groups. For this study, raw scores for the pre-test and post-test were compared with the norms for
349 children aged 5;8-5;11 and raw scores for follow-up were compared to the norms for children aged
350 6;0-6;5 (van Haaften et al., 2019).

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

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

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

369 2018). The Figure Weights task was used in the baseline ~~and~~, weekly testing, and follow-up testing,
370 alongside the MDT. Reliability and validity of the WISC-V-NL are adequate (Wechsler, 2018).

371

372 *Insert table 3 around here*

373

374 Analysis

375 Kendall's tau test was used to test for change on the MDT measures CV, CVC, CCVC, CVCC,
376 accuracy, consistency, fluency, place, manner and vowel. The level of significance was set at $p < 0.05$.

377 Kendall's tau test was also used to analyze ~~the~~ changes in the non-verbal control task Figure Weights.

378 The CAI and CAT ~~are norm-referenced. These~~ norms were used to describe changes on these

379 measures. ~~Norm groups in CAI are divided into age groups that span four months for the younger~~

380 ~~groups and six months for the two oldest groups. For this study this means that raw scores for pre-test~~

381 ~~and post-test were compared with the norm group for children aged 5;8-5;11 and raw scores for~~

382 ~~follow-up were compared to the norm group for children aged 6;0-6;5 (van Haaften et al., 2019).~~

383

384 Results

385 The participant in this study received 20 30-minute SMTA sessions, over a period of 11 weeks instead
386 of 10, due to one week of illness during the treatment period. The number of ~~(partial) items~~ items (both

387 full items at the sentence level and parts of items at the word level) that was trained in each session

388 varied from four to seven. The SLP and MT implemented ~~repetition~~ repetitions of items and breaks as

389 needed, resulting in different numbers of items practiced across sessions. In one session there were

390 four trained items, in ~~4~~ twelve sessions there were five, in five sessions there were ~~6~~ six trained items

391 and in two sessions there were seven. Each week, the items trained in the first session were repeated

392 in the second. The first week, the more personally ~~motivated~~ motivating item with the name of his

393 stuffed animal was trained, building from practicing words to the full sentence. In the second and third

394 week, items with the persistently difficult /r/ and /h/ were practiced at the word and sentence level. In

395 the fourth through sixth week, ~~the~~ items with consonant clusters were trained at the syllable, word and

396 ~~sentences~~ sentence levels. From week seven on, focus was placed on ~~the~~ multisyllabic words, as well

397 as consonant clusters, building up to sentence level for items containing such words. Nine out of ten of

398 the formulated targets at sentence level were trained. The boy was able to produce the fourth item

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

416

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

420

421 *Insert table 4 around here.*

422

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

428 Improvement ~~foron~~ PVC and the production of initial clusters ~~that, which~~ was obtained after treatment,
429 was not maintained at follow-up.

430

431

Insert table 5 around here.

432 Tasks and measures ~~off~~from the CAI showed varying results. The CAI does not provide critical
433 differences, therefore a change of $Z > + 0.5$ was set as the norm for clinically relevant change. Z-
434 scores were calculated using the means and SDs published by Van Haaften et al. (2019). In Picture
435 Naming, presented in figure 1, there was clinically relevant change for PCCI over the study period ($Z +$
436 0.51 at follow-up compared to pre-test), but no clinically relevant changes for PVC ($Z + 0.42$ over
437 treatment period). The production of initial clusters in Picture Naming improved after treatment ($Z +$
438 5.46), and the gains were mostly maintained at follow-up ($Z + 4.29$). Results for cluster reductions in
439 the same task, for which scores were inverted to reflect that a higher score means ~~less~~fewer cluster
440 reductions, showed clinically relevant change over the treatment period, reaching ceiling level after
441 treatment ($Z + 0.97$), which was maintained at follow-up ($Z + 1.16$). ~~In~~On Non-Word Imitation,
442 presented in figure 2, results for PCCI showed clinically relevant change after treatment ($Z + 1.22$), but
443 this was not maintained at follow-up ($Z + 0.16$). Results for PVC showed an increase from below
444 average to within normal limits directly after treatment ($Z + 0.85$), but this improvement was not
445 maintained at follow-up ($Z - 1.0$). The production of initial clusters in Non-Word Imitation did not
446 change directly after therapy, but did improve at follow-up ($Z + 1.34$). Results for cluster reduction in
447 this task showed clinically relevant change, reaching ceiling level after treatment ($Z + 2.8$), which was
448 maintained at follow-up ($Z + 2.83$). Results for consistency are presented in figure 3. Word consistency
449 increased directly after treatment ($Z + 0.80$), but this improvement was not maintained at follow-up.
450 Non-word consistency did not increase directly after treatment, but improvement was apparent at
451 follow-up ($Z + 1.71$). The DDK ~~of~~task from the CAI, which measures maximum repetition rate (MRR;
452 syllables per second) showed no change for the sequential items /pa/, /ta/ and /ka/. Results for the
453 alternating items are presented in figure 4. For the items /pata/ and /taka/, results were lower directly
454 after treatment ($Z - 0.48$ and $Z - 2.42$) and at follow-up ($Z - 0.16$ and $Z - 0.69$). For /pataka/ the
455 scores improved after treatment ($Z + 3.59$) from not being able to perform this sequence at pre-test to
456 within normal limits at follow-up ($Z + 4.21$).

457

Insert figures 1,2, 3 and 4 around here

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

461 *Insert table 6 around here*

462

463 Results on several measures of the MDT showed change after treatment and over the study period.
464 For syllable structure, there was a significant gradual improvement on CV (Kendall $\tau = 0.633$, $p < .01$)
465 and CCVC (Kendall $\tau = 0.396$, $p < .05$) structures as shown in figure 5. Results for the structures CVC
466 (Kendall $\tau = 0.290$, $p > .05$) and CVCC (Kendall $\tau = 0.132$, $p > .05$) showed no significant change over
467 the study period. Significant gradual improvement was found ~~in~~for the measures of accuracy (Kendall
468 $\tau = 0.433$, $p < .05$) and consistency (Kendall $\tau = 0.447$, $p < .05$), but not for fluency (Kendall $\tau = 0.211$,
469 $p > .05$) as shown in figure 6. Figure 7 shows significant gradual improvement for the measures place
470 (Kendall $\tau = 0.513$, $p < .01$), manner (Kendall $\tau = 0.656$, $p < .01$) and vowel (Kendall $\tau = 0.356$, $p <$
471 $.05$). ~~On~~For the scores on the non-verbal control task Figure Weights, which is also shown in figure 7,
472 there ~~is~~was no significant improvement over the study period (Kendall $\tau = -0.210$, $p > .05$).

473

474 *Insert figure 5, 6 and 7 around here*

475

476 **Discussion**

477

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

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

490 The study was designed to evaluate SMTA on a range of outcome measures, including intelligibility in
491 daily communication and communication attitude, as well as a variety of measures of speech motor
492 planning and programming in specific speech-~~of speech~~-motor tasks. Tasks included picture naming,
493 non-word imitation, word- and non-~~wordrepetition~~word repetition, and DDK₁ with outcome measures
494 such as percentage of initial consonants correct, production of clusters, consistency and fluency.

495 Overall results showed progress ~~in~~on measures of speech motor planning and programming occurring
496 directly after treatment. Improvement ~~of~~on intelligibility and communication attitude was mostly
497 obtained over the entire study period and became apparent at follow-up.

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

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

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

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

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

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

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

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

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

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

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

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

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

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

604

605 Limitations and directions for future research

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

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

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

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

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

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

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

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

669

670 **Conclusion**

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

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

682

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688

689 **Data access statement**

690 The data can be obtained by contacting the first author.

691

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851

852 **Appendix A: Phonetic repertoire**

853

854 **Appendix B: Medical history of the participant**

855

856 **Appendix C: Trained items**

857

858 **Appendix D: Items and scoring of the Modified Diadochokinesis Test**

859

860 **Figures**

861 **Figure 1. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and Clred in Picture**

862 **Naming.** PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels

863 Correct, CCVC = Percentage of Initial Clusters Correct, Clred = cluster reduction. T1 = pre-test, T2 =

864 post-test, T3 = follow-up (after two months of no treatment), ★ = $Z > + 0.5$, clinically relevant change.

865

866

867 **Figure 2. Raw scores and Z-scores for measures of PCCI, PVC, CCVC and Clred in**

868 **~~Nonword~~Non-word Imitation.** PCCI = Percentage Consonants Correct In Initial position, PVC =

869 Percentage of Vowels Correct, CCVC = Percentage of Initial Clusters Correct, Clred = cluster

870 reduction. T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = $Z > +$

871 0.5, clinically relevant change.

872

873 **Figure 3. Raw scores and Z-scores for measures of consistency in word and ~~nonword~~non-word**

874 **repetition.** WR = word repetition, NWR = Nonword repetition, T1 = pre-test, T2 = post-test, T3 =

875 follow-up (after two months of no treatment), ★ = $Z > + 0.5$, clinically relevant change.

876

877 **Figure 4. Raw scores and Z-scores for maximum repetition rate in alternating sequences.** T1 =

878 pre-test, T2 = post-test, T3 = follow-up (after two months of no treatment), ★ = $Z > + 0.5$, clinically

879 relevant change.

880

881 **Figure 5. Scores for syllable structure on the MDT.** Wk = week, B = baseline, T = treatment, ★ =

882 significant $p < .05$, Kendall's tau test.

883

884 **Figure 6. Scores for Consistency, Accuracy and Fluency on the MDT.** Wk = week, B = baseline, T

885 = treatment, ★ = significant $p < .05$, Kendall's tau test.

886

887 **Figure 7. Scores for Place, Manner and Vowel on the MDT and the control task Figure Weights.**

888 Wk = week, B = baseline, T = treatment, ★ = significant $p < .05$, Kendall's tau test.

889

891 **Table 1. Structure of a 30 minute treatment session**
 892

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

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

896 **Table 2. Structure of aan SMTA exercise**
 897

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 (<u>direct imitation</u>)	Child + SLP	5 repetitions of target item*
Semi-spontaneous speaking (respond to question)	Child + SLP	3 repetitions of target item*

898 SLP = speech and language pathologist, MT = music therapist. ^At the introduction the child is invited
 899 to participate when they are able to repeat the target item correctly. *The number of repetitions is
 900 influenced by the length of the target item. A melody usually contains 8 repetitions for words and 4
 901 repetitions for sentences. The melody is sung and chanted at least two times. The numbers here are
 902 the minimal number of repetitions that are achieved during one exercise.
 903

904 **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

905 ICS-Dutch = Intelligibility in Context Scale – Dutch (McLeod et al., 2012a), FAN = Phonological
 906 Analysis for Dutch (Beers, 1995), CAI = Computer Articulation Instrument (Maassen et al., 2019), CAT
 907 = Communication Attitude Test (Brutten & Vanryckeghem, 2003), MDT = Modified Diadochokinesis
 908 Test (Hurkmans et al., 2012), Figure ~~Weighs~~Weights from the Wechsler Intelligence Scale for
 909 Children, ~~vijfde editie; Nederlandstalige bewerking~~Fifth Edition; Dutch Edition (Wechsler, 2018). Full
 910 descriptions of these measures are provided under ‘Outcome measures’ in the ~~method~~methods
 911 section.
 912

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

ICS-Dutch	T1	T2	T3
Father	3 _{T2} .7	3 _{T2} .4	3 _{T2} .9
Mother	3 _{T2} .6	4	4 _{T2} .1
Parents combined	3 _{T2} .6	3 _{T2} .7	4

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

915

916 **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

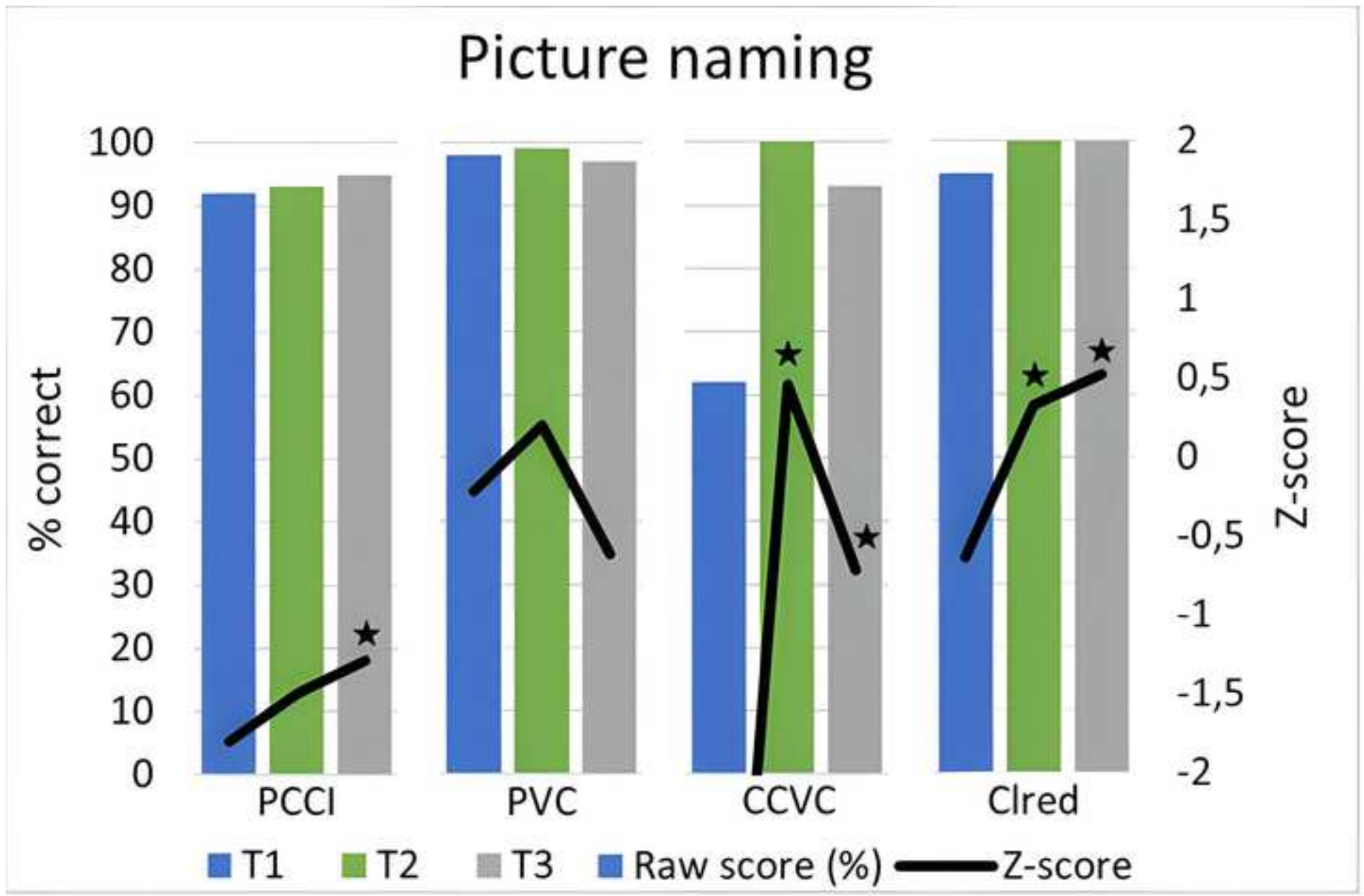
917 PCCI = Percentage Consonants Correct In Initial position, PVC = Percentage of Vowels Correct,
918 CCVC = Percentage of Initial Clusters Correct. T1 = pre-test, T2 = post-test, T3 = ~~Follow~~Follow-up
919 (after two months of no treatment).
920

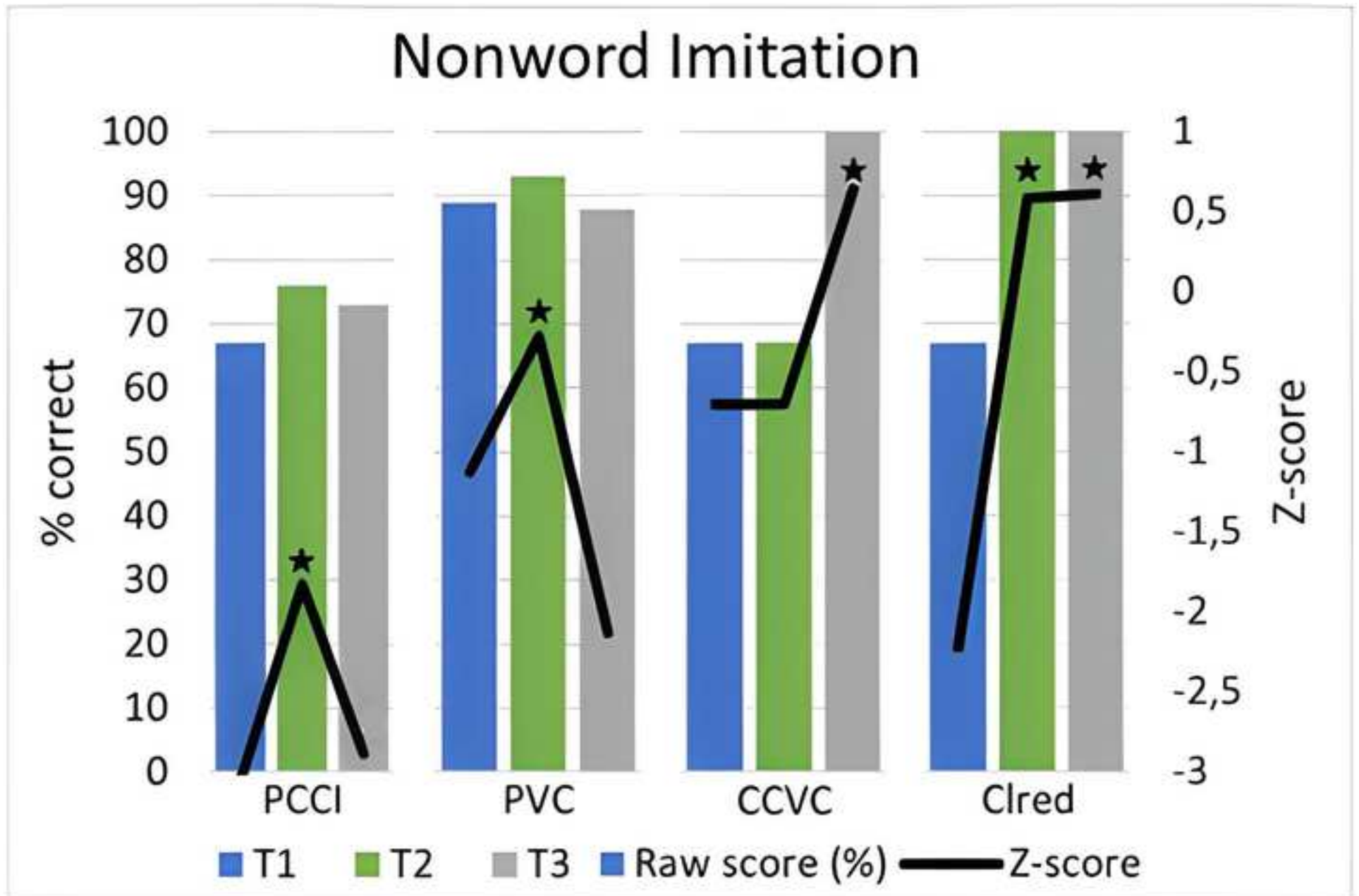
921 **Table 6. Results for the CAT.**

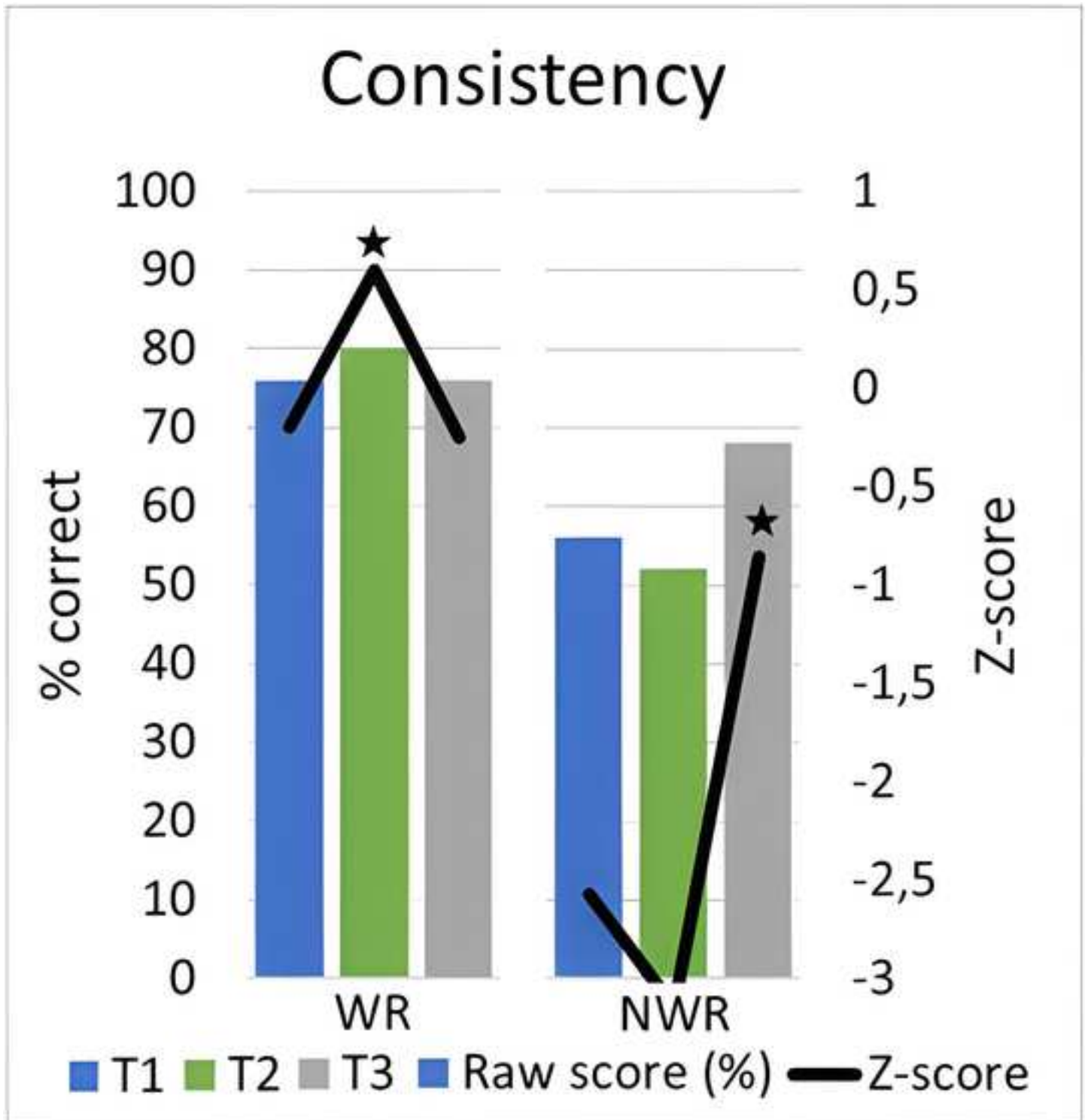
CAT	T1 (Form A)	T2 (Form B)	T3 (Form B)
Raw scores	7	7	3
SD	+ 0.25	+ 0.43	- 0.52

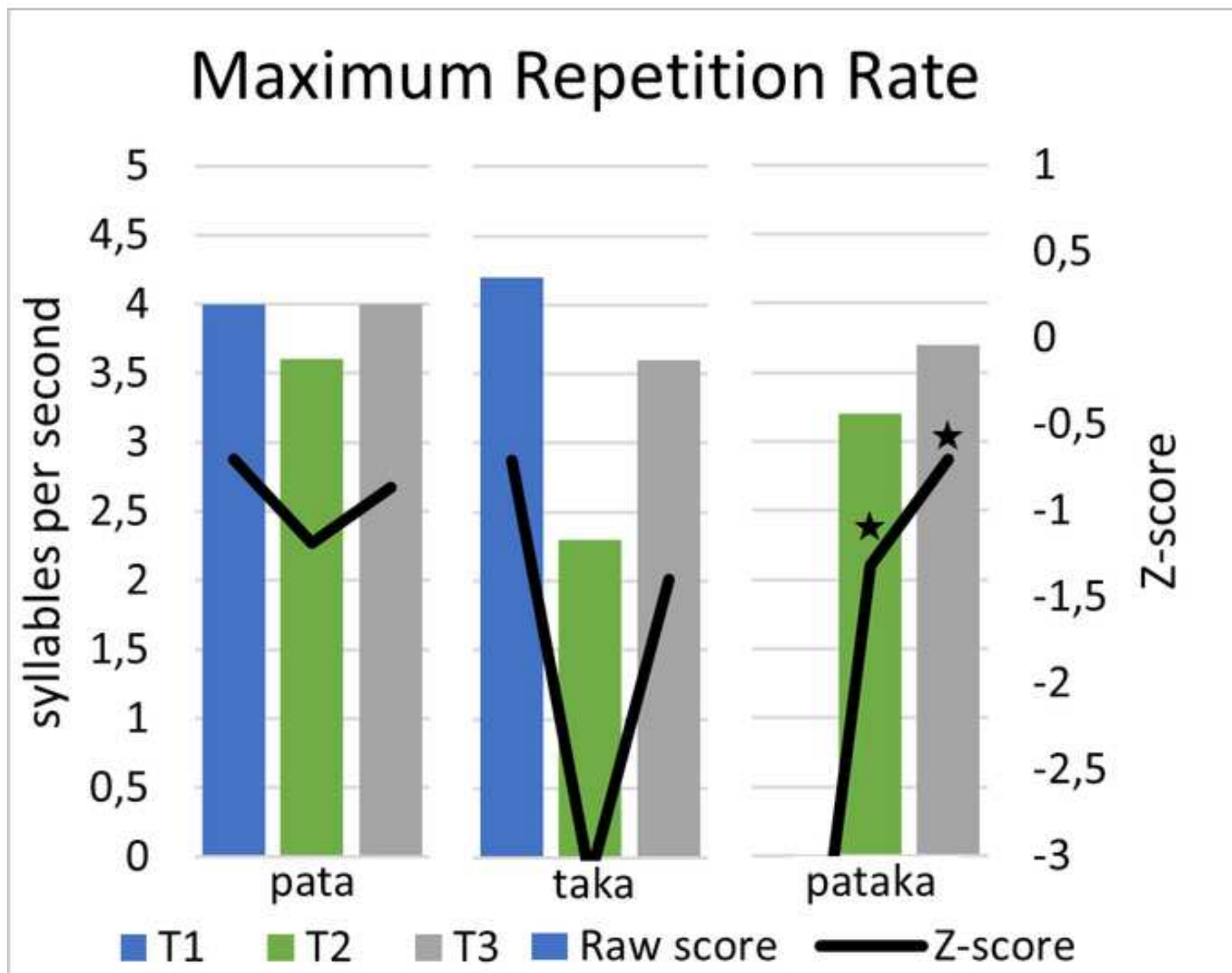
922 SD = standard deviation, T1 = pre-test, T2 = post-test, T3 = follow-up (after two months of no
 923 treatment).

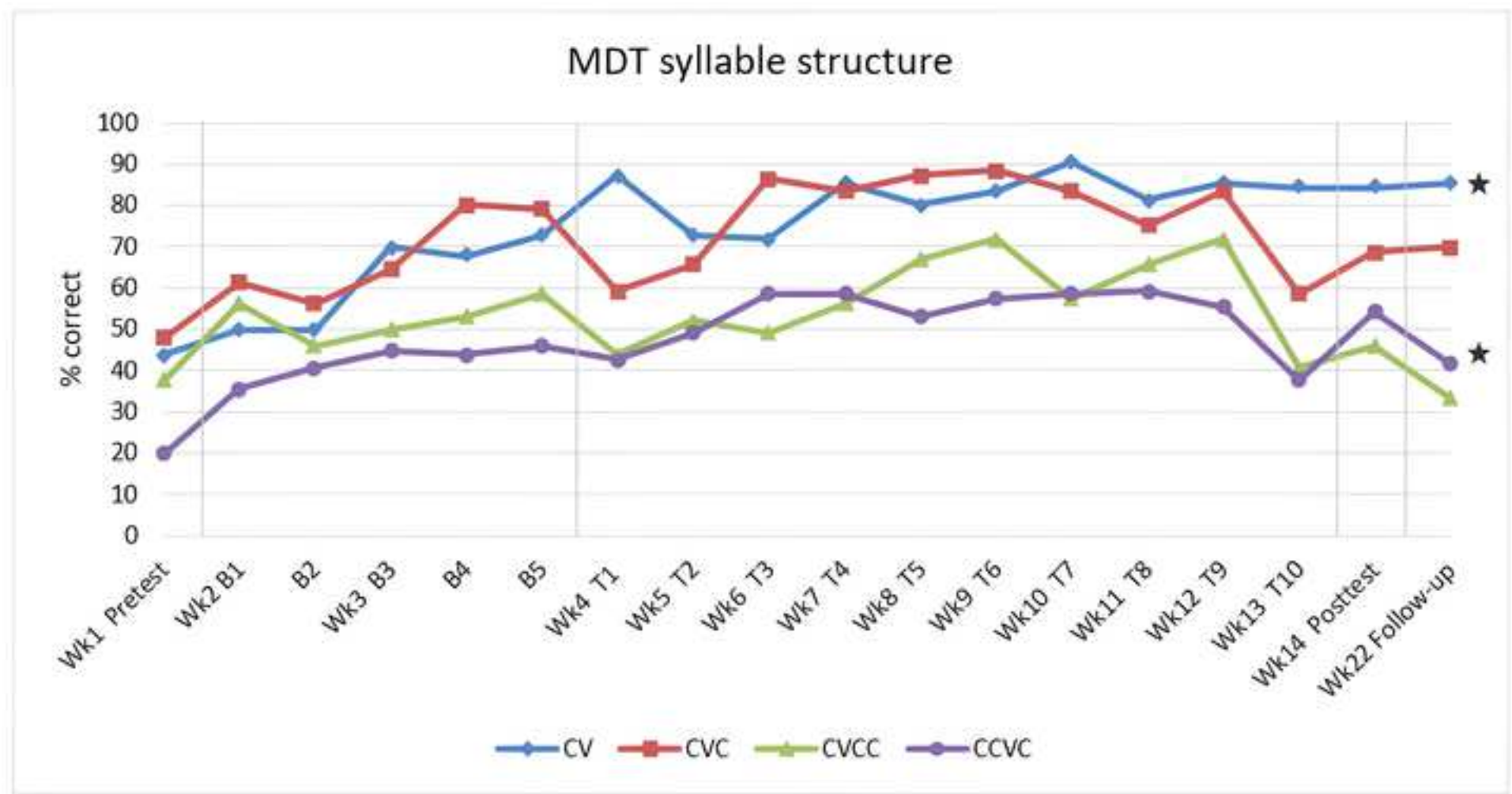
924 Note: A higher score indicates a more negative attitude towards their own speech.

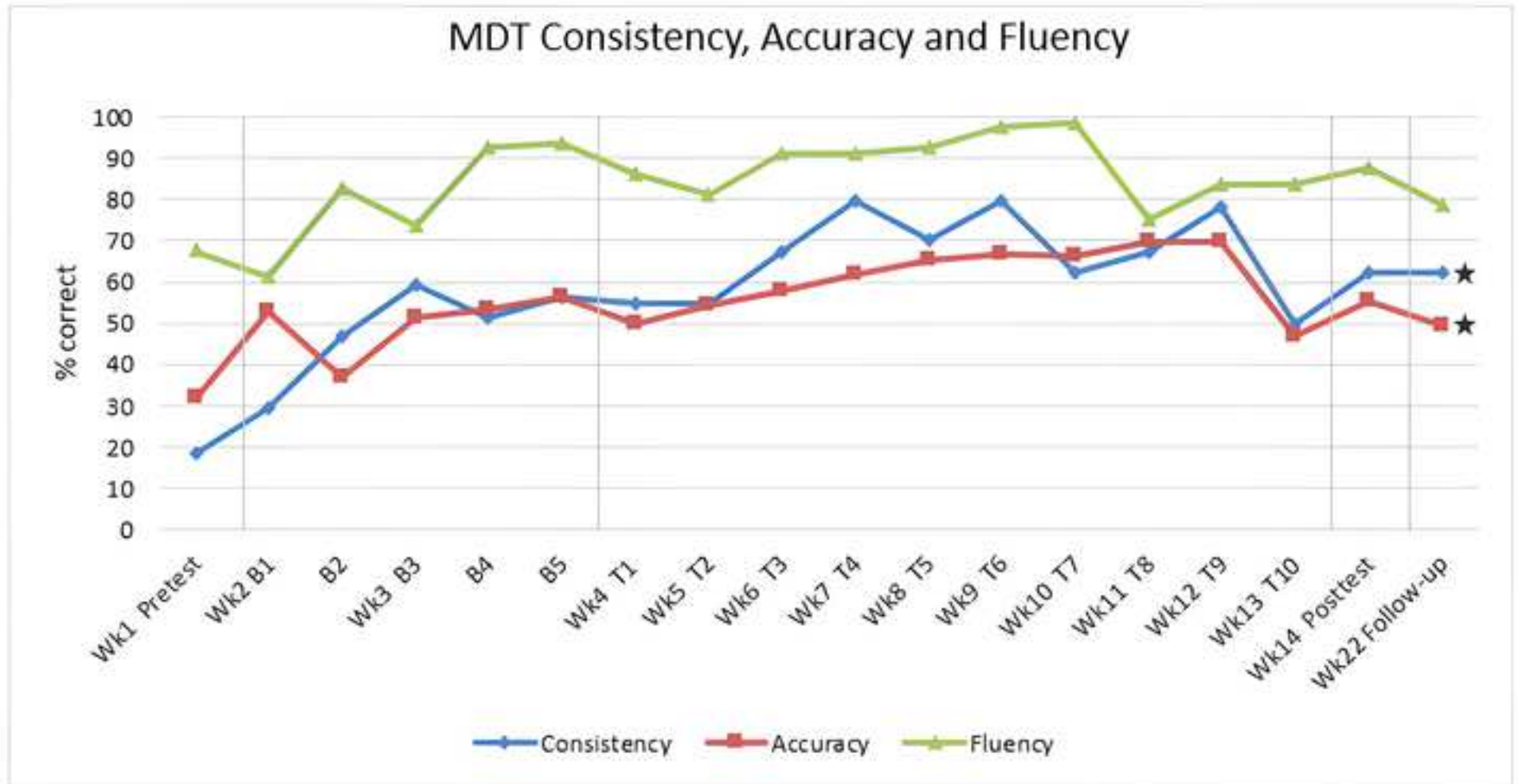


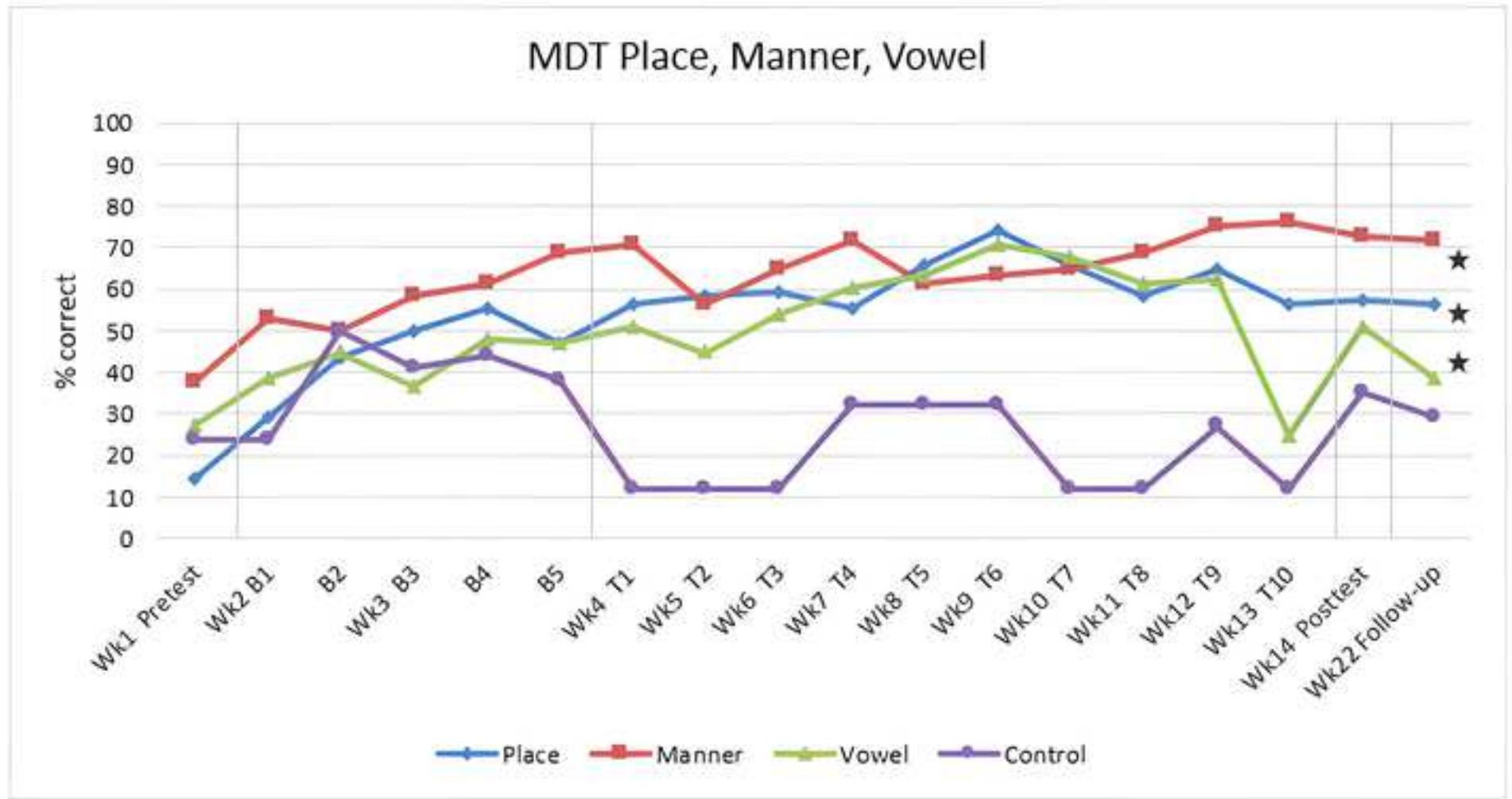












Reviewers comments

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 personally 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.

The results are clear, overall. However, there is contradictory information about whether or not the child made progress on consonants, in lines 585-593

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.

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.

Table 2: Change title to "Structure of an SMTA exercise

Add parenthetical explanation to "Alternately speaking": (direct imitation)

Table 3: Correct spelling of "Weights". If possible, translate the Wechsler citation into English. Change penultimate word to "methods" instead of "method."

Tables 4 & 6: Please use periods in decimals instead of commas, which would be confusing for many readers.

Table 5: Fix spelling of "Follow-up"

6. Discussion/Conclusions The discussion has been improved markedly.

Although it is much improved, I still have many suggestions about the writing, listed below by line:

113-114: Change to: "They also use rate/rhythm control strategies.."

Thank you.

We changed this section to reflect that there were a few consonants that showed improvement, thus leading to three categories of consonants in the analysis of spontaneous speech.

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.

Thank you for pointing out this mistake. This asterisk is now deleted from the figure.

We changed this as suggested.

We added this as suggested.

We applied these changes as suggested.

We changed this as suggested.

We fixed this.

Thank you.

Thank you for your suggestions to improve the writing. We responded to them below.

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 Repetition..”	We changed this as suggested.
386: “Additionally, the occurrence...”	We changed this as suggested.
387: “cluster reduction (CIRed)..”	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 separately 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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Appendix

SMT in the treatment of CAS - Appendix A.docx

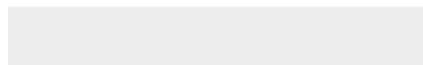




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Appendix

SMT in the treatment of CAS - Appendix B.docx

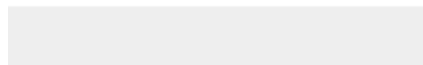




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Appendix

SMT in the treatment of CAS - Appendix C.docx





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Appendix

SMT in the treatment of CAS - Appendix D.docx

