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1	Auditory-Motor Mapping Training Facilitates Speech and Word Learning in Tone-
2	Language-Speaking Children with Autism: An Early Efficacy Study
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12 13 14 15 16 17 18	E-mail addresses: chenfeianthony@gmail.com Conflict of Interest: The authors have declared that no competing interests existed at the time of publication. Funding Statement: This work was in part supported by the Major Program of National Social Science Foundation of China (18ZDA293), the Natural Science Foundation of China overseas collaboration grant (31728009), the General Research Fund of the Research Grants Council of

20 Abstract

Purpose: It has been reported that tone-language-speaking children with autism demonstrate speechspecific lexical tone processing difficulty although they have intact or even better-than-normal processing of nonspeech/melodic pitch analogues. In this early efficacy study, we evaluated the therapeutic potential of an Auditory-Motor Mapping Training (AMMT) in facilitating speech and word output for Mandarinspeaking nonverbal and low-verbal children with autism, in comparison with a matched non-AMMT-based control treatment.

Method: Fifteen Mandarin-speaking nonverbal and low-verbal ASD children participated and completed all the AMMT-based treatment sessions by intoning (singing) and tapping the target words delivered via an app, while another fifteen participants received control treatment. Generalized linear mixed-effects models were created to evaluate the speech production accuracy and word production intelligibility across different groups and conditions.

Results: Results showed that the AMMT-based treatment provided a more effective training approach in accelerating the rate of speech (especially lexical tone) and word learning in the trained items. More importantly, the enhanced training efficacy on lexical tone acquisition remained at two weeks post-therapy, and generalized to untrained tones that were not practiced. Furthermore, the low-verbal participants showed higher improvement compared to the nonverbal participants.

37 Conclusions: These data provide the first empirical evidence for adopting the AMMT-based training to
38 facilitate speech and word learning in Mandarin-speaking nonverbal and low-verbal children with autism.
39 This early efficacy study holds promise for improving lexical tone production in Mandarin-speaking
40 children with autism but should be further replicated in larger-scale randomized studies.

41 Keywords: autism, nonverbal, low-verbal, Mandarin, lexical tones, Melodic Intonation Therapy

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Auditory-Motor Mapping Training Facilitates Speech and Word Learning in Tone-Language-Speaking Children with Autism: An Early Efficacy Study

44

45 **1 Introduction**

Autism spectrum disorder (ASD) is a neurodevelopmental disorder identified by a constellation of early-appearing social communication deficits and restricted, repetitive sensorymotor behaviours (American Psychiatric Association, 2013). The delayed/atypical language development – historically linked to a defining feature – has been removed from the diagnostic criteria and is now regarded as one of the co-occurring characteristics of autism. The goal of this study is to integrate recent advances in speech therapy to facilitate speech and word learning in tone-language-speaking children with ASD.

53 Nonverbal and Minimally Verbal Children with ASD and Behavioral Interventions

Estimates vary, with 25%-46% of children with ASD reported as minimally verbal past 54 the age of 5 years (Norrelgen et al., 2015; Rose et al., 2016; Tager-Flusberg & Kasari, 2013), 55 meaning that they have an expressive vocabulary fewer than 20 (Kasari et al., 2013), or 30 56 intelligible words (Bal et al., 2016; Plesa-Skwerer et al., 2016). Besides, some individuals with 57 58 ASD even remain nonverbal with a complete absence of functional speech, and lack the ability to communicate with others using spoken language (Klinger et al., 2002; Koegel et al., 2009; 59 60 Turner et al., 2006). While children with ASD showed deficits in social communication and 61 social interaction, this is further exacerbated in nonverbal and minimally verbal children with ASD due to their severely delayed speech development. Increased speech and word output is 62 63 considered a positive prognostic indicator of outcomes for nonverbal or minimally verbal 64 children with ASD (Lord et al., 2006). Recently, the growing availability of computers and

smartphones/tablets has generated a great deal of enthusiasm for their potential to help
ameliorate speech deficits in minimally-verbal children with ASD, such as the computer-assisted
3-D virtual pronunciation tutor (Chen et al., 2019), and the Proloquo2Go iPad app (King et al.,
2014). In consideration of autistic features especially among nonverbal and low-verbal
individuals who begin treatment with limited or no spoken words, available behavioral
interventions often tried to present speech sounds with computerized "high-tech" solutions
(Tager-Flusberg & Kasari, 2013).

72 Mandarin Chinese as a Tonal Language and the State of the ASD Research in China

73 A very recent review estimated that ASD prevalence in China was comparable to the Western world (about 108 per 10,000) using standardized case identification protocol (Sun et al., 74 2019). However, the situation of ASD in China lags considerably behind those in the West in 75 terms of public awareness, education opportunities, and life outcomes of people with autism (Liu 76 et al., 2016; Yu et al., 2020). It is compelling to come up with a language-specific training 77 78 approach for children with ASD who live in a country hosting nearly 20% of the world's population. Modern Mandarin is a tone language with a relatively simple syllable structure, 79 consisting of initials (consonants), finals (including monophthong, diphthong, triphthong, or 80 81 nasal finals with a vowel nucleus), as well as lexical tones (Chen et al., 2017). Specifically, Mandarin Chinese is the official and widely spoken language in China and is also used in some 82 83 other countries/regions, which phonologically differs from English. For example, Mandarin is a 84 syllable-timed tone language that exploits variations in both pitch height and pitch direction at the syllable level to distinguish lexical meanings (Figure 1), while English is a stress-timed non-85 86 tone language (Mok & Dellwo, 2008). There are four citation tones in Mandarin Chinese: high-87 level Tone 1 (T1, [55]), mid-rising Tone 2 (T2, [35]), low-falling-rising Tone 3 (T3, [214]), and

88	high-falling Tone 4 (T4, [51]). These four lexical tones are essential elements of Mandarin
89	speech sounds, and are used to differentiate lexical meanings. For instance, "i" spoken with the
90	four distinct tones can respectively mean "doctor" (T1), "move" (T2), "rely on" (T3), and "easy"
91	(T4). Thus, changing the lexical tone in a tone language has the same kind of effect as changing
92	a vowel or a consonant. In contrast, variation in pitch contours in non-tone languages mainly
93	conveys different moods or intonations without changing the word content (Wang, 1973). Thus,
94	speech therapy in tone-language-speaking children with ASD should not only aim at enhancing
95	consonant and vowel production (segmental elements of speech), but additionally, also aim at
96	improving lexical tone production (supra-segmental element of speech).

- 97
- 98

[Figure 1 about here]

99

100 Superior Music and Nonspeech Processing but Impaired Lexical Tone Processing in ASD

Music is one of the most meaningful and popular forms of nonspeech sound; like speech, 101 it has developed to take advantage of the efficiencies of the human auditory system (Baldwin, 102 2012). In the research into auditory processing, several studies demonstrated that children with 103 104 ASD prefer musical or nonspeech stimuli over speech and attend to them more (Dawson et al., 1998; Kuhl et al., 2005). Many children with ASD showed enhanced capacity of music processing 105 and exhibited strong interest in learning and making music (Buday, 1995; Hairston, 1990; Heaton 106 107 et al., 1998). Irrespective of the tone inventory (Mandarin or Cantonese), the enhanced or at least 108 preserved non-linguistic pitch perception skill in ASD generalized to those from a tone language background (Cheng et al., 2017; Wang et al., 2017; Yu et al., 2015). Nevertheless, such an 109 110 enhanced acoustic pitch perception skill was conversely changed to the compromised perception

of lexical tones when the pitch changes are phonologically relevant for the tone language speakers 111 with ASD (Chen et al., 2016; Lau et al., 2020; Wang et al., 2017; Yu et al., 2015). In terms of 112 speech production, when compared with age-matched typically-developing (TD) children, the 113 ASD group aged 3–6 years showed an apparent speech delay in the production of Mandarin initials, 114 finals, as well as lexical tones (Wu et al., 2020). Since both music notes and lexical tones share the 115 116 same psychoacoustical attribute of pitch information, it would be reasonable to take advantage of the relative strength of musical skills to compensate for the relative weakness of speech sound, 117 especially lexical tone acquisition, for tone-language-speaking children with ASD. 118

119 The Intonation-Based Interventions in ASD

Given the behavioral resemblance between singing and speaking as well as neural 120 121 overlap in responses to speech and musical stimuli (Peretz et al., 2015), researchers have begun to examine the therapeutic effects of singing/intonation, and how it can potentially ameliorate 122 123 some of the speech deficits associated with neurological disorder such as ASD (Wan et al., 2010). Two earlier case studies have described the positive role of singing (intoned rather than 124 spoken verbal stimulus) in facilitating speech development of children with autism (Hoelzley, 125 1993; Miller & Toca, 1979). Recently, Melodic Intonation Therapy (MIT; Albert et al., 1973; 126 Sparks et al., 1974), initially designed for improving spoken language in left-hemisphere stroke 127 patients with severe nonfluent aphasia, has been introduced into speech therapy in ASD. The 128 MIT approach involves the musical elements of both melody and rhythm through the use of 129 pitched vocalization or singing in combination with left-hand rhythmic tapping to provide cueing 130 for syllable production (Norton et al., 2009). An modified version of MIT, called Auditory-131 132 Motor Mapping Training (AMMT), was initially proposed by Wan et al. (2011) to facilitate speech output for English-speaking nonverbal children with autism. The intonation-based 133

AMMT combines intonation (singing) of bi-syllabic words or phrases and the use of a pair of 134 tuned drums to activate bimanual motor activities. All six English-speaking nonverbal children 135 with autism who participated in Wan et al.'s (2011) study showed noticeable improvements in 136 their ability to articulate several word approximations. Furthermore, the efficacy of AMMT has 137 been further corroborated in English-speaking minimally verbal children with autism 138 139 (Chenausky et al., 2016) and one more-verbal child with autism (Chenausky et al., 2017) when compared with a control treatment. In this study, we apply and assess the effect of intonation-140 based AMMT, with proven efficacy as an intervention for English-speaking children with ASD 141 (Chenausky et al., 2016, 2017; Sandiford et al., 2013; Wan et al., 2011), on Mandarin-speaking 142 children with ASD. 143

To this end, the present study evaluated the therapeutic potential of AMMT in facilitating 144 speech output for tone-language-speaking children with ASD. Moreover, as suggested by Wang 145 (1978), children do not learn speech by acquiring units like phonemes or allophones, but rather by 146 147 gradually adding lexical items to their repertoire. Two important studies (Ferguson & Farwell, 1975; Hsieh, 1972) investigated the development of phonological production in relation to the 148 acquisition of words, and showed that there was a primacy of word learning during speech 149 150 development. Thus, in this study, our intervention based on a smartphone/iPad app called Music-Mediated and Lexicon-Integrated (MMLI) training, tries to combine speech and word learning. 151 The current training study aimed to assess the efficacy of AMMT-based MMLI in comparison to 152 153 a matched non-AMMT-based control treatment, Speech Repetition Therapy (SRT) (Chenausky et al., 2016). Specifically, this study aimed to address the following research questions: 154

155	a) Over the intensive treatment sessions of MMLI, would the nonverbal and low	-verbal
156	Mandarin-speaking children with ASD show any improvement in speech and	l word
157	productions?	
158	b) Compared with the control group, would the MMLI group show a greater improve	nent in
159	the speech production accuracy of initials, finals, and lexical tones, as well as th	e word
160	production intelligibility?	
161	c) If significant improvements were observed in MMLI, could the benefits be retained	ed after
162	the cessation of daily treatment sessions and generalize to untrained items?	
163		
164	2 Methods	
165	2.1 Participants	
166	Upon entering the treatment sessions, 30 participants with autism were randomly	
167	assigned to one of two treatment groups. The MMLI group ($n = 15$, two girls) represented	the
168	experimental group, and the SRT group ($n = 15$, two girls) acted as the active control group	p.
169	They were recruited from Cangzhou Research Centre for Child Language Rehabilitation.	
170	Permission to conduct this study was obtained from by the local institutional review board	of the
171	Hong Kong Polytechnic University, ensuring appropriate adherence to informed consent	
172	procedures. The parents provided informed, written permission for their children to partici	pate
173	before any assessments were administered.	
174	The clinical diagnosis of ASD was established according to the DSM-5 criteria for	ASD
175	(American Psychiatric Association, 2013), and further confirmed (16 out of 30 participant	5)
176	using the Autism Diagnostic Observation Schedule-2 (ADOS-2; Lord et al., 2012) by	
177	pediatricians and child psychiatrists in child hospitals. The clinical diagnosis of ADOS-2 v	vas not
178	conducted at the time of data collection, and the average time between the study procedure	s and

the diagnosis was 8.14 months (SD = 5.29 months). Since the Mandarin version of ADOS-2 has 179 not been officially validated and widely adopted in China (Sun et al. 2013; Yu et al., 2015), for 180 those cases where an ADOS report was absent (14 out of 30 participants), we confirmed 181 diagnoses using the Chinese version of the Gilliam Autism Rating Scale-Second Edition 182 (GARS-2; Gilliam, 2006) under the supervision of a licensed clinical psychologist. Other 183 184 inclusion criteria were: (1) the ability to imitate two piano notes; (2) the ability to sit in a chair and take part in instructed activities for around 15 minutes at a time; (3) the ability to imitate 185 gross motor activities such as clapping hands, and imitate oral motor movements; (4) not having 186 the following comorbidities: cerebral palsy or tuberous sclerosis, hearing/sight impairment, 187 Down's syndrome, uncontrolled seizures, and organic impairment of oral or laryngeal structures 188 (Chenausky et al., 2016; Wan et al., 2012). An additional 11 children with autism were found to 189 be ineligible and were excluded from the current study because they either did not meet the four 190 inclusion criteria or could not regularly attend all the required treatment sessions. 191 192 Furthermore, 30 TD children were recruited from one local kindergarten to investigate the speech, language and cognitive levels of our participants with autism before training in 193 reference to age-matched neuro-typical ones. The TD children did not participate in the training 194 195 sessions. Although the TD children were not specially screened for autism symptoms via standardized instruments, they met none of the diagnostic DSM-5 criteria for ASD from an 196 197 interview with their parents or teachers. As shown in Table 1, the participants with ASD did 198 show significant speech, language and cognitive delays compared to age-matched TD children. The children with autism were divided into two subgroups in the current study: Nonverbal status 199 200 was defined as having complete absence of intelligible words before training; low-verbal status 201 was defined as using expressive vocabulary of no more than 50 words, based on parent reports as

202	well as the pretest measures. The term "low-verbal" was used in several previous studies (Yoder
203	& Stone, 2006; Kasari et al., 2008) to describe individuals with ASD with severe speech,
204	language and cognitive delays.
205	
206	[Table 1 about here]
207	
208	2.2 Study Design
209	2.2.1 Language and Cognitive Evaluation
210	A randomized control design was used in an effort to determine the effectiveness of the
211	experimental treatment (MMLI) and control for various external factors. Prior to training, all
212	participants were firstly assessed with their language ability (Ning, 2013), then nonverbal IQ
213	using the Primary Test of Nonverbal Intelligence (PTONI, Ehrler & McGhee, 2008), and then
214	working memory (Millman & Mattys, 2017). The two treatment groups did not differ from each
215	other in terms of chronological age, language ability, nonverbal IQ, as well as working memory
216	(Table 2).
217	
218	[Table 2 about here]
219	
220	Language Ability: The overall language ability (Chen et al., 2017; Ning, 2013) was
221	evaluated for each Mandarin-speaking participant, which consists of five subtests (including Test
222	of Mandarin Grammar, Word Definition Test, Rapid Automatized Naming, Narrative Test, and
223	Sentence Comprehension Test). These subtests evaluated both language comprehension and

language expression, and aimed to assess different aspects of language abilities such as
phonology, lexicons, grammar, and semantics. The administration time is around 30 minutes.

Primary Test of Nonverbal Intelligence: PTONI (Ehrler & McGhee, 2008) is a
theoretically sound, research-based method of assessing reasoning abilities in young children
aged from 3:0 to 9:11. The PTONI was normed on a culturally diverse demographic sample from
diverse language backgrounds. Testing takes approximately 15 minutes.

Working Memory: In order to evaluate the short-term phonological working memory
(Millman & Mattys, 2017), we administered a forward digit span task, In the task, a series of
numbers were played to participants auditorily and they were asked to repeat them immediately.
For each digit length (two to nine digits), there were two separate items. The response for each
item was regarded as correct and awarded 0.5 point only when the participants could correctly
repeat every digit in the right order. The full score for the test of forward digit span is 8.

236

237 2.2.2 Probe Assessments

238 For low-verbal participants, probe assessments were conducted in the Pretest (Test 1), Midtest (Test 2), Immediate Posttest (Test 3), and Delayed Posttest (Test 4). After the first 12 239 240 treatment sessions, three nonverbal children with ASD from each treatment group (six in total) demonstrated no progress at all, and still remained nonverbal. They received the second-round 241 12 treatment sessions in the same modality as the first 12 sessions, with 24 treatment sessions in 242 total (Figure 4). The change in protocol was not determined a priori. We decided to conduct 243 more training sessions for these nonverbal participants, in an effort to figure out whether the null 244 training efficacy was due to the relatively short-term training duration or due to the failure of the 245

current approach. For those six nonverbal participants, probe assessments were performed not 246 only in Tests 1–4, but also in the second-round Midtest (Test 5), and Immediate Posttest (Test 6). 247 During each probe assessment, the same picture naming task was utilized, with trained 248 (Set 1) and untrained (Set 2) picture stimuli intermixed and presented in random order. Set 1 249 consisted of 60 lexical items that were presented during probe assessments and also practiced 250 251 during therapy sessions. Set 2 included 12 high-frequency items, which were not practiced during treatment, but presented during the probes (see Supplemental Material S1). The untrained 252 stimuli in Set 2 were used to assess the transfer of learning to untrained stimuli. The children 253 254 were encouraged to make more than one attempt (at most three times in total) if they did not respond, or failed to produce the target word correctly, in an effort to guarantee that the 255 spontaneous productions could truly reflect production capacity in children with autism. 256 Researchers who administered the probe assessments were blind to treatment condition. 257 However, it should be noted that during the probe assessments, no correct demonstrations were 258 provided. The cueing sentence is "WHAT IS THIS?" without any information about the naming 259 of the target pictures. The spontaneous productions from each child were recorded in a sound-260 isolated room for further analyses. As shown in Table 2, the two treatment groups did not differ 261 262 from each other in the production accuracy of speech sounds (initials, finals, and tones) and word production intelligibility in the Pretest (i.e., at baseline assessment). 263

264

2.2.3 The Development of a Smartphone/iPad App

265

The app was developed based on the Ionic Framework

266 (<u>https://ionicframework.com/docs</u>), which uses a user interface (UI) toolkit for building mobile

and desktop apps via web technologies. It is an open-source front-end framework for HTML5

hybrid mobile application to help developers use the same source code to generate app files for

269	both Android and iOS-based platforms. The primary function of the current app is to assist
270	children's speech learning as well as word learning. The home page presented six themes,
271	including vegetables, fruits, animals, daily necessities, snacks, and toys (Figure 2a). Under each
272	theme, a total of 10 high-frequency lexical items were chosen, resulting in 60 trained lexical
273	items in total. These 60 trained words were disyllabic nouns relevant to children's early-acquired
274	vocabulary. These trained items contained all the 21 initials, 39 finals, four citation tones as well
275	as neutral tone, and T3 tone sandhi in Mandarin phonology. The untrained words included 12
276	high-frequency lexical items, which included early-developing ([m, n, t, t ^h]), middle-developing
277	$([k, k^h, t\epsilon^h, \epsilon])$, and late-developing Mandarin consonants ([ts, ts^h, s, tş, tş^h, ş]), monophthongs
278	([u,], y]), diphthongs ([uA, Gu, ou]), triphthongs ([iGu, iou]), finals with a nasal coda ([an, iaŋ,
279	υŋ, iŋ, əŋ]), as well as all the tonal categories in Mandarin. Please refer to Supplemental Material
280	S1 for details.
281	
282	[Figure 2 about here]
283	
284	Then, for each word item, one corresponding picture was presented in the center of the
285	interface as the visual cue, and two piano icons on the left and right sides (Figure 2b). The
286	natural speech sounds of 60 trained words (120 syllables) were recorded from one female
287	announcer with standard Mandarin pronunciation. To construct the piano-timbre nonspeech
288	sounds, first, a piano note (C4) of 261 Hz frequency was created, then the level pitch tier was
289	replaced with the pitch contour extracted from each syllable (120 syllables in total) using the
	replaced with the pitch contour extracted from each synaple (120 synaples in total) using the
290	Pitch-Synchronous Overlap Add implanted in Praat (Boersma & Weenink, 2016). In this way,

the piano-timbre nonspeech sounds share the same pitch contours as those in natural speech

292	sounds (Figure 3). All the piano-timbre nonspeech sounds were normalized to 500 ms, and
293	equally for root-mean-square intensity level, at 70 dB SPL. During MMLI training, when tapping
294	the icons from left to right, the piano-timbre nonspeech sounds that match the pitch contours of
295	natural lexical tones for the first and second syllables would be played, one tap per syllable. As
296	for the source code, please refer to https://github.com/introfei/VoiceTrain and for compiling,
297	packaging, and uploading issues, please refer to https://github.com/introfei/Blog/issues/3 to see
298	more details.
299	[Figure 3 about here]
300	
301	2.2.4 Treatment Protocol
302	Therapists were female undergraduate students majoring in speech rehabilitation from the
303	Cangzhou Research Centre for Child Language Rehabilitation, and were trained specifically to
304	provide both MMLI and SRT training. If one therapist taught a child participant in the MMLI
305	group, she would need to teach another child participant in the SRT group. They had an average
306	of 1.73 years of experience in teaching children with autism prior to this study. Therapists firstly
307	learned from the first author on how to perform and follow the treatment protocol. They were
308	also required to make a practice of teaching other children with autism who were not
309	participating in this study until they were familiar with the whole protocol. The treatment
310	protocol of MMLI and SRT is shown in Table 3. They were conducted with intensive repetition
311	in a highly structured environment. The SRT is designed to be similar in several respects to
312	conventional forms of speech therapy, while lacking the key elements of MIT (Chenausky et al.,
313	2016). While SRT also presents verbal stimuli through the app interface and contains the same
314	steps and speech outputs as MMLI, in SRT the verbal stimuli are spoken, not intoned; and there

315	is no bimanual hand tapping on piano icons. The video demonstrations of one trial for the two
316	groups (MMLI and SRT) have been shown in Supplemental Material S2. To monitor the
317	treatment fidelity, all treatment sessions were videotaped to evaluate therapists' adherence to the
318	protocol. We reviewed five videotaped sessions (41.67 % of all treatment sessions) selected at
319	random from each child (75 treatment sessions for the MMLI group, and 75 treatment sessions
320	for the SRT control group). Each treatment session contains 30 trials (10 trials x 3 repetitions),
321	and each trial contains five steps (Table 3). On all the 2,250 MMLI trials, therapists intoned the
322	target word and tapped the piano icons, and neither of the 2,250 SRT trials were intoned or
323	tapped. Furthermore, over a total of 11,250 steps assessed in MMLI group, 91 (0.81 %) had
324	repeated steps and 44 (0.39%) had omitted steps. Over the total 11,250 steps assessed in SRT
325	group, 103 (0.92 %) had repeated steps and 38 (0.34%) had omitted steps.
326	
327	[Table 3 about here]
328	
329	2.2.5 Training Procedure
330	The therapy sessions were conducted in clinical treatment rooms at Cangzhou Research
331	Centre for Child Language Rehabilitation. The child participants in both MMLI and SRT groups
332	received short-term intensive training – 12 treatment sessions 6 times per week, over a 2-week
333	period. However, after the first round of training, three nonverbal children with ASD (out of the
334	15 participants) from each treatment group demonstrated no progress at all, and still remained

- nonverbal. These six nonverbal participants received the second-round 12 treatment sessions,
- with 24 treatment sessions in total (Figure 4). Each treatment session began with a warm-up
- stage, followed by 10 lexical trials (each trial was repeated three times) of one specific theme.

338	The three repetitions of each trial were blocked together, and all the five steps of each trial were
339	repeated. The sequence of steps was the same and strictly followed the protocol regardless of the
340	response from the child (i.e., if the child did not respond or did not pronounce the word
341	correctly). Each session lasted about 50-60 minutes, including breaks, which occurred every ten
342	to fifteen minutes, based on the child's stamina. The order of six training themes within one
343	intervention phase was randomized using a Latin Square among participants. The training order
344	in the second phase is a repetition of that in the first intervention phase for each child. While
345	receiving MMLI or SRT, the child participants were not allowed to engage in any other speech
346	therapy activities in regular school programs. As required in the consent form, parents agreed to
347	withhold other interventions while testing an unproven intervention in order to satisfy scientific
348	objectives.
349	
373	
350	[Figure 4 about here]
350 351	[Figure 4 about here]
350 351 352	[Figure 4 about here] 2.3 Outcome Measures
350 351 352 353	[Figure 4 about here] 2.3 Outcome Measures 2.3.1 Speech and Word Production Measures
 350 351 352 353 354 	[Figure 4 about here] 2.3 Outcome Measures 2.3.1 Speech and Word Production Measures The recorded productions were transcribed offline by Mandarin-speaking transcribers
 350 351 352 353 354 355 	[Figure 4 about here] 2.3 Outcome Measures 2.3.1 Speech and Word Production Measures The recorded productions were transcribed offline by Mandarin-speaking transcribers who were totally blind to the current study design in order to minimize experimental bias. Before
 350 351 352 353 354 355 356 	[Figure 4 about here] 2.3 Outcome Measures 2.3.1 Speech and Word Production Measures The recorded productions were transcribed offline by Mandarin-speaking transcribers who were totally blind to the current study design in order to minimize experimental bias. Before transcription, an expert majoring in phonetics picked out the best sample from each child's
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 350 351 352 353 354 355 356 357 358 	[Figure 4 about here] 2.3 Outcome Measures 2.3.1 Speech and Word Production Measures The recorded productions were transcribed offline by Mandarin-speaking transcribers who were totally blind to the current study design in order to minimize experimental bias. Before transcription, an expert majoring in phonetics picked out the best sample from each child's utterances if more than one attempt was produced for one specific target word. The criterion is to choose the one with higher mean accuracy in the productions of initial, final, and lexical tone. If
 350 351 352 353 354 355 356 357 358 359 	[Figure 4 about here] 2.3 Outcome Measures 2.3.1 Speech and Word Production Measures The recorded productions were transcribed offline by Mandarin-speaking transcribers who were totally blind to the current study design in order to minimize experimental bias. Before transcription, an expert majoring in phonetics picked out the best sample from each child's utterances if more than one attempt was produced for one specific target word. The criterion is to choose the one with higher mean accuracy in the productions of initial, final, and lexical tone. If the nonspeech-like vocalization was produced for a certain target, the nonspeech-like
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across all the probe assessments. The transcribers were told about the corresponding
requirements in advance, but without being told about the nature and purpose of the current
study. The transcribers needed to transcribe all the 3,634 words (7,268 syllables) and they were
allowed to listen to the sound stimuli as many times as they wanted until they were confident to
make a transcription. The order of presentation for the transcription task was randomized. Each
sound stimulus was transcribed once by each transcriber.

First, for speech production measure, the disyllabic word was split into two syllables, 368 which were transcribed separately. The 7,268 produced syllables were randomized and further 369 370 transcribed using the International Phonetic Alphabet (IPA) by another five trained experts majoring in linguistics (mean age = 24.50 years). Especially for the tonal coding, the exact tonal 371 categories were chosen from the following descriptions: high-level tone (T1), mid-rising tone 372 (T2 or full sandhi: *T3 [35]), dipping tone (typical T3), high-falling tone (T4), low-falling tone 373 (half-T3: *T3 [21]), and the neutral tone. Such stringent measures of phonetic transcription using 374 375 IPA were aimed to assess the actual correct production of phonemes (Munro & Derwing, 1995) in a more fine-grained and precise manner. If none of the Mandarin phonology matched the 376 transcription, the transcribers would log "none" instead. Each coder needed around 40 hours in 377 378 total to complete the transcription of all the initials, finals, and tones. Three outcome measures were included to evaluate the speech production accuracy: Percent Initials Correct, Percent 379 Finals Correct, and Percent Tones Correct, which were calculated with the number of correctly 380 381 transcribed initials, finals, and lexical tones divided by the total number of syllables, respectively. 382

Second, in terms of word production measure, the 3,634 produced words were
randomized and presented to five native speakers of Mandarin not majoring in linguistics (mean

age = 21.38 years) with the E-Prime 2.0 program (Psychology Software Tools Inc., USA). The 385 transcribers were not told about the target words before transcription. They were asked to write 386 down each word they heard with two Chinese characters (each Chinese character representing a 387 morpheme in the Mandarin word) one by one in a spreadsheet to evaluate word production 388 intelligibility (i.e., how well the word is understood regardless of if all phonemes were accurately 389 390 produced; transcribers were allowed to take their best guess). When none of the Chinese characters were suitable to be used to make a transcription, transcribers would log "none" 391 instead. Each coder needed around 15 hours to complete the transcription of words. The number 392 of correctly coded characters (morphemes) was divided by the total number of characters 393 (morphemes) to yield "Percent Morphemes Correct". 394

To assess agreement among five raters, Kendall's Concordance Coefficient W was calculated in this study (Legendre, 2005). The interrater reliability with Kendall's coefficients of 0.802 for initial transcription, 0.793 for final transcription, 0.828 for tone transcription, and 0.884 for morpheme coding was reached, exhibiting relatively high inter-rater reliability.

399 **2.3.2** User Experience

The user experience evaluation (Chen et al., 2019) was executed after the completion of 400 401 all the treatment sessions, which was rated using a 5-point Likert scale based on the child's training performance (5 – the highest degree, 1 – the lowest degree). Each corresponding 402 403 therapist rated the user experience for her own student. Such subjective observations evaluated 404 the ways in which children with ASD approached different training methods. The user experience evaluation included five aspects: enjoyment, cooperation, consistency, interest, and 405 motivation. Enjoyment refers to the degree of apparent pleasure in the learning process; 406 407 cooperation means the degree of collaboration in learning a trial (whether the child could follow

all the five steps within a trial); consistency indicates the continuity of the overall coordination
throughout the learning process; interest refers to the degree of apparent interest in the training
materials; motivation represents the degree of the apparent initiative before treatment sessions
(whether the child appears to want to participate in training). The operational definitions of five
aspects are shown in Supplemental Material S3.

413 **2.4 Statistical Analyses**

All the statistical analyses of outcome measures were performed in R (R Core Team, 414 2014). For the analyses of production accuracy, the generalized linear mixed-effects models 415 (GLMMs) were created using the lme4 package (Bates et al., 2014). It is feasible for GLMM in 416 R to include all the item responses transcribed from five different transcribers, and have them 417 calculated within a single statistical model. In each GLMM, treatment group (MMLI vs. SRT), 418 test, and their two-way interaction acting as fixed effects. When fitting GLMMs, participant and 419 *item* were included as random effects. The R code for the full model: Accuracy \sim treatment 420 421 group * test + (1 + test | participant: treatment group) + (1 + treatment group * test | item). Byparticipant and by-item random intercepts and random slopes for all possible fixed factors were 422 423 included in the full model (Barr et al., 2013), which was compared with a simplified model that 424 excluded a specific fixed factor using the ANOVA function in ImerTest package (Kuznetsova et 425 al., 2017). Moreover, post-hoc pairwise comparisons were calculated with the lsmeans package 426 (Lenth, 2016) with Tukey adjustment.

For the analyses of user experience, a generalized Poisson regression model (Consul & Famoye, 1992) was constructed in R using a glmer function (family = 'poisson'), with *treatment group* (MMLI vs. SRT), *aspect* (motivation, consistency, interest, cooperation, and enjoyment), and their two-way interaction acting as fixed effects. The generalized Poisson regression model

is often a first-choice model for counts-based datasets, and has been found useful in fitting overdispersed as well as under-dispersed count data. Given that the nonverbal and low-verbal
participants in this study received different amounts of treatment sessions, their results were
reported separately.

435

436 **3 Results**

437 **3.1 Outcomes in Nonverbal Participants**

There were six nonverbal participants with ASD (G101, G102, and G103 in MMLI 438 group; G201, G202, and G203 in SRT group), who received 24 treatment sessions across four 439 intervention phases in total (Figure 4). The probe assessment data were collected six times 440 before, during, and after therapy. Only one participant with autism from the MMLI group (G103) 441 began to acquire some initials, finals, lexical tones, as well as words in the trained items during 442 the second-round training, while all the other five participants remained nonverbal even after 24 443 444 treatment sessions. Furthermore, none of the six nonverbal participants showed any improvement in the untrained items after training. For the evaluation of user experience, the subject from the 445 MMLI group (G103) who showed gains in speech and word learning of trained items also 446 obtained relatively higher scores of user experience, especially in the aspect of enjoyment. 447

448

449 **3.2 Outcomes in Low-Verbal Participants**

In total, there were 24 low-verbal participants with ASD (n = 12 in each treatment group) who received 12 treatment sessions across two intervention phases (Figure 4). Figure 5 shows the percentage of correct productions from two treatment groups in both trained and untrained items. The x-axis represents the probe assessment sessions and the y-axis stands for the

454	percentage of correct initials, finals, lexical tones, and morphemes, respectively, from left to
455	right.
456	[Figure 5 about here]
457	
458	3.2.1 Production Accuracy of Initials
459	The GLMM was performed on the production accuracy of initials in trained stimuli, and
460	the statistical results only showed a significant main effect of <i>test</i> (χ^2 (3) = 1143.70, <i>p</i> < .001).
461	However, the GLMM did not reveal the significant main effect of <i>treatment group</i> (χ^2 (1) = 1.33,
462	$p = .468$) nor the interaction effect of <i>treatment group</i> * <i>test</i> (χ^2 (3) = 3.35, $p = .341$). Further
463	examination on the effect of test implied that the low-verbal participants from both groups
464	showed significant improvement in producing initials in the trained items (all $ps < .001$), at
465	Midtest, Immediate Posttest, as well as at Delayed Posttest. Furthermore, the results showed that
466	the treatment methodology (MMLI vs. SRT) did not lead to outcome differences in the
467	production accuracy of initials in the trained stimuli across all the probe assessments (Figure 5a
468	& Table 4a).
469	
470	[Table 4 about here]
471	
472	Then, the GLMM on the accuracy of initials in the untrained items also merely showed a
473	main effect of <i>test</i> (χ^2 (3) = 56.01, <i>p</i> < .001), while the main effect of <i>treatment group</i> (χ^2 (1) =
474	0.05, $p = .818$) and the interaction of <i>treatment group</i> × <i>test</i> (χ^2 (3) = 0.51, $p = .918$) were not
475	significant. For both MMLI and SRT groups, as shown in Figure 5b, the number of correctly
476	produced initials of untrained stimuli increased significantly after the whole 12 treatment

sessions at Immediate Posttest ($\beta = -0.39$, SE = 0.07, t = -6.03, p < .001) and at follow-up 477 assessment at Delayed Posttest ($\beta = -0.34$, SE = 0.07, t = -5.22, p < .001). Moreover, the two 478 groups performed similarly in the production accuracy of initials in the untrained items across all 479 the probe assessments (Figure 5b & Table 4b). 480

481

3.2.2 Production Accuracy of Finals

482 For the trained items in Set 1, the GLMM model on the accuracy of finals showed a significant main effect of test (χ^2 (3) = 1192.78, p < .001), while the main effect of treatment 483 group ($\chi^2(1) = 0.03$, p = .861) was not significant. There was a significant two-way interaction 484 of *treatment group* × *test* (χ^2 (3) = 17.61, *p* < .001). Compared with Pretest, both treatment 485 groups made significant progress in the trained finals over the course of treatment (all ps < .001). 486 Furthermore, the two training groups differed after two intervention phases (Figure 5a & Table 487 5a), with the MMLI group showing a higher production accuracy of trained finals than the 488 matched control group at Immediate Posttest ($\beta = 0.18$, SE = 0.04, t = 4.88, p < .001), while no 489 group differences were found at the other probe assessments (all ps > .05). 490

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- 492

[Table 5 about here]

493

494 For the untrained items in Set 2 (Figure 5b), the GLMM on production accuracy of finals only reveal a main effect of test ($\chi^2(3) = 79.11$, p < .001). Neither the main effect of treatment 495 group ($\chi^2(1) = 0.18$, p = .675) nor the interaction of treatment group × test ($\chi^2(3) = 3.06$, p496 = .382) was significant. The post hoc analyses demonstrated that both treatment groups showed 497 significant progress in number of correctly produced finals in untrained items only at the 498 immediate posttest ($\beta = -0.59$, SE = 0.07, t = -7.97, p < .001). Moreover, the two groups 499

performed similarly in the production accuracy of finals in the untrained items (Figure 5b &Table 5b).

502

503 3.2.3 Production Accuracy of Tones

The GLMM on the accuracy of trained tones showed significant main effects of 504 *treatment group* (χ^2 (1) = 334.59, p < .001) and *test* (χ^2 (3) = 1493.43, p < .001), as well as a 505 significant two-way interaction of *treatment group* × *test* (χ^2 (3) = 112.87, p < .001). In 506 comparison to the performance in Pretest (Figure 5a & Table 6a), both MMLI and SRT groups 507 showed significant improvement at tone production of the trained items over the course of 508 treatment (all ps < .001). Nevertheless, the growth rate was quite different, with participants who 509 received MMLI training showing much higher production accuracies of trained tones compared 510 with those receiving SRT, at Midtest ($\beta = 0.22$, SE = 0.04, t = 5.62, p < .001), Immediate Posttest 511 $(\beta = 0.61, SE = 0.04, t = 16.26, p < .001)$, as well as Delayed Posttest ($\beta = 0.45, SE = 0.04, t =$ 512 12.11, *p* < .001). 513 514 [Table 6 about here] 515 516 Then, GLMM was performed on the accuracy of tones in untrained items, and the 517

statistical results exhibited significant main effects of *treatment group* (χ^2 (1) = 40.85, p < .001) and *test* (χ^2 (3) = 85.20, p < .001), as well as a significant interaction of *treatment group* × *test* (χ^2 (3) = 14.27, p < .01). The low-verbal participants from the MMLI group had significant progress in production of lexical tones in untrained items, at Immediate Posttest and Delayed Posttest (ps < .001), while those from the SRT group showed no progress over the course of treatment (all ps > .05). In terms of group difference (Figure 5b & Table 6b), the experimental group of MMLI obtained much higher accuracy of tones in untrained stimuli after 12 treatment sessions at Immediate Posttest ($\beta = 0.42$, SE = 0.12, t = 3.41, p < .001) and two weeks later at Delayed Posttest ($\beta = 0.49$, SE = 0.13, t = 3.90, p < .001).

527

3.2.4 Word Production Intelligibility

528 First, for the trained items, the GLMM on word production intelligibility (Percent Morphemes Correct) showed significant main effects of *treatment group* (χ^2 (1) = 33.22, p 529 < .001) and test (χ^2 (3) = 1611.81, p < .001), as well as a significant interaction of treatment 530 group × test (χ^2 (3) = 38.65, p < .001), indicating that the two training groups showed different 531 trajectories of word learning in the trained items. As shown in Figure 5a, compared to the 532 baseline performance in Pretest, both MMLI and SRT groups showed noticeable improvements 533 in trained word production (all ps < .001) when tested at Midtest, Immediate Posttest, and 534 follow-up assessment two weeks later. In terms of group difference at different timepoints (Table 535 7a), the two treatment groups performed similarly on Percent Morphemes Correct in the Pretest 536 $(\beta = -0.015, SE = 0.038, t = -0.40, p = .688)$ and Midtest $(\beta = 0.011, SE = 0.036, t = 0.29, p)$ 537 = .772), whereas the MMLI group produced higher accuracy in the trained morphemes than the 538 control SRT group in the Immediate Posttest ($\beta = 0.247$, SE = 0.035, t = 7.03, p < .001) as well 539 as Delayed Posttest ($\beta = 0.103$, SE = 0.035, t = 2.94, p < .01). 540

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- 542

[Table 7 about here]

543

544 Second, for the untrained items, the GLMM on word production intelligibility (Percent 545 Morphemes Correct) revealed a significant main effect of *test* (χ^2 (3) = 128.84, *p* < .001), but the

main effect of *treatment group* did not reach significance ($\chi^2(1) = 0.07$, p = .789). There was a 546 significant interaction of *treatment group* × *test* (χ^2 (3) = 19.09, *p* < .001). Post-hoc analysis 547 showed that compared to the performance in Pretest, the MMLI group produced more untrained 548 morphemes correctly in Midtest, Immediate Posttest, and Delayed Posttest (ps < .05). Over the 549 same probe assessments, however, the SRT group only produced more untrained morphemes in 550 the Immediate Posttest ($\beta = -0.54$, SE = 0.10, t = -5.37, p < .001) right after 12 treatment sessions 551 (Figure 5b & Table 7b). For the between-group difference, across all the probe assessments, the 552 MMLI group performed similarly to the control SRT group in terms of % Morphemes Correct in 553 554 the untrained words (all ps > .05).

555 **3.2.5** User experience

The generalized Poisson regression model on scores of user experience for the low-verbal children with ASD did not show significant main effects of *aspect* (χ^2 (4) = 8.22, *p* = .084) and *treatment group* (χ^2 (1) = 3.02, *p* = .082). Moreover, the Poisson regression model did not reveal the significant interaction of *treatment group* × *aspect* (χ^2 (4) = 0.24, *p* = .993). Thus, despite the trend, the experimental group of MMLI (*M_{MMLI}* = 3.42) did not receive higher scores across different aspects of user experience compared with the control group of SRT (*M_{SRT}* = 2.65).

563 **4 Discussion**

The current MMLI training app aimed to facilitate speech and word learning in tonelanguage-speaking children with ASD. For the low-verbal participants, while both MMLI and SRT groups were found to be effective in enhancing production skills, results suggested a faster rate of improvement in the production of finals, lexical tones, and words in the trained items for the experimental MMLI group. Moreover, the advantage of MMLI training transferred to the untrained items in terms of lexical tone production. For the six nonverbal participants, however,
only one from the MMLI group responded to treatment in the trained items, while others from
both training groups showed no progress and remained nonverbal even after 24 treatment

- 572 sessions. We will discuss these findings hereunder.
- 573

4.1 Potential Mechanisms Responsible for the Training Efficacy of MMLI

574 MMLI resulted in greater improvements than SRT in most of the outcome measures for the Mandarin-speaking participants, which largely corroborated the efficacy of the AMMT-based 575 training approach, with well-proven efficacy as a treatment for English-speaking children with 576 577 autism (Chenausky et al., 2016, 2017; Sandiford et al., 2013; Wan et al., 2011). The data reported here further proved that the two key elements of MIT - intonation and hand tapping -578 added greatly to MMLI's effectiveness in children from another language system. When tapping 579 on the virtual piano presented through the app, the nonspeech sounds with piano timbre would be 580 generated in an effort to mimic the music-making activities. In contrast with previous studies 581 582 (Chenausky et al., 2016, 2017; Sandiford et al., 2013; Wan et al., 2011), our MMLI system did not utilize real musical notes, but was modified to match various pitch variations of lexical tones 583 in Mandarin phonology. Hoelzley (1993) proposes that the unique timbre of musical instruments 584 585 may increase motivation and attention in children with autism. The MMLI group seemed to enjoy the treatment more than the SRT group based on the clinical observation, but it was not 586 587 supported by the statistics.

588 Furthermore, music making through bimanual tapping on the tuned piano icons is a 589 multimodal activity that not only captures the attention in children with autism, but also possibly 590 primes and integrates the bilateral sensorimotor networks with shared motor, auditory, and visual 591 neural representations of the articulatory/hand movements (Bangert et al., 2006; Lahav et al.,

2007). In particular, it is speculative that the arcuate fasciculus (AF), a fiber bundle that connects 592 593 the auditory perceptual regions in the temporal lobe with the motor-related regions in the frontal lobe (Catani et al., 2005), might be developed or reconstructed through intonation and music-594 making activities (Wan et al., 2010, 2011). The AF was thought to play an important role in 595 auditory-motor mapping (Chenausky et al., 2017), and might be responsible for the bidirectional 596 597 mapping between speech articulation and acoustics (Leclercq et al., 2010), as well as facilitating new word learning especially in the left bundle (López-Barroso et al., 2013). Another neural 598 substrate likely to be engaged during music making is the putative mirror neuron system (MNS). 599 600 It has been suggested that dysfunctional MNS underlies some of the speech and language deficits in individuals with ASD (Iacoboni & Dapretto, 2006). The elements in MMLI training, such as 601 imitation, hand tapping, and synchronization, might activate brain regions that overlap with 602 MNS, thus highlighting the potential benefits of such sensorimotor training to facilitate 603 expressive language in developmental disorder such as autism (Overy & Molnar-Szakacs, 2009). 604

4.2 Training Efficacy for Low-Verbal Children with ASD

While MMLI holds promise for improving speech learning in Mandarin-speaking 606 children with ASD in general, the effectiveness of MMLI was unbalanced among different 607 608 components of syllables (i.e., initials, finals, and tones). A shown in Figure 5a, low-verbal participants with ASD receiving MMLI started to show superiority over those receiving SRT in 609 610 their ability to correctly articulate Mandarin lexical tones in trained items as early as Midtest, and 611 such advantage further expanded after 12 treatment sessions at Immediate Posttest and was maintained at Delayed Posttest. For speech production of Mandarin finals which use vowel(s) as 612 613 the whole final or as the nucleus, the low-verbal MMLI participants only experienced 614 comparatively greater improvement than the SRT participants after 12 treatment sessions, and

such advantage did not persist at the follow-up assessment. In terms of the speech production of 615 616 Mandarin initials that were composed of consonants, the two treatment groups performed similarly in the trained items over all the probe assessments. Furthermore, for the untrained items 617 (Figure 5b), MMLI produced significantly greater gains merely in the lexical tone learning in 618 low-verbal children with ASD compared to the control therapy, SRT. Such generalization skills 619 620 would be greatly beneficial to children with ASD, who show difficulty in transferring learned knowledge to a new context (Church et al., 2015; Happé & Frith, 2006). In a short conclusion, as 621 observed from the current data, the efficacy of MMLI was much higher in the training of lexical 622 623 tones, followed by vowels, and then consonants.

The greater improvement on lexical tone acquisition should not be surprising given that 624 relative to SRT, MMLI presented participants with additional information of pitch contours 625 embedded in piano-timbre nonspeech. Individuals with ASD have often demonstrated pitch or 626 melodic processing superiority in various musical and nonspeech stimuli (e.g. Gomot et al., 627 2002; Heaton, 2005; O'Riordan & Passetti, 2006). Accumulating evidence pointed to a two-way 628 transferability of pitch expertise across domains of music and speech (as lexical tones) in neuro-629 typical children and adults (Bidelman et al., 2013; Nan et al., 2018; Wong et al., 2007). By 630 631 targeting the clinical population, the current data provided the first empirical evidence of using the relative strength of music, a ubiquitous nonspeech form, to compensate for the relative 632 633 weakness of speech sounds, especially lexical tone acquisition for tone-language-speaking 634 children with ASD. One recent study (Nan et al., 2018) demonstrated that the six months of piano training not only enhanced the lexical tone discrimination, but also improved vowel and 635 636 consonant discrimination in 4- to 5-year-old Mandarin-speaking TD children, suggesting 637 strengthened common sound processing across domains underlying the benefits of musical

training. In this training study, however, we failed to detect the benefits of music-supported 638 639 MMLI training on the acquisition of initials (consonants). On the one hand, the shorter, weaker, and more aperiodic consonants in speech sounds are likely to be impacted more in a co-occurring 640 nonspeech background than the stable, periodic components of tones and vowels. On the other 641 hand, since the syllable-initial consonants were acquired later than the vowels and tones in 642 643 Mandarin-speaking TD children (Hua & Dodd, 2000), the relatively short-term training duration in this study may be another potential factor leading to the failure of transfer effects on the late-644 645 acquired consonant production.

646 4.3 Training Efficacy for Nonverbal Children with ASD

The nonverbal participants in our study belong to the subgroup of the autism spectrum 647 with severe language/cognitive impairment. They were completely nonverbal, despite having 648 received extensive speech therapy (four to 16 months) prior to recruitment. In the current study, 649 except for one nonverbal child with ASD responding to the MMLI treatment, the other five 650 nonverbal participants could not correctly produce even one trained word after 24 treatment 651 sessions, and meanwhile, they received lower scores on user experience. It is unlikely that this is 652 due to the stringent measure of phonetic transcription, since these five nonverbal participants did 653 654 not even spontaneously produce any verbal attempts during probe assessments. In contrast, as reported in Wan et al. (2011), the English-speaking nonverbal participants with autism who 655 656 received similar AMMT training began to vocalize some "word approximations" after 10-15 657 treatment sessions. It should be noted that the speech samples produced by the nonverbal participants in Wan et al.'s (2011) study were imitations rather than spontaneous productions, 658 659 and should inform the degree and nature of progress. In our study, however, the spontaneous 660 speech samples were collected from participants in a picture naming task without cueing or

demonstration. Another possibility is that the nonverbal participants in our study were more severely impaired in terms of language and cognitive capacity compared to those in Wan et al.'s (2011) study. Given the extreme challenges these participants face, more treatment sessions or some other training approaches should be delivered to nonverbal children with ASD. In some cases, both parents and therapists have observed an increase in speechlike vocalizations during vocal play in daily life, which might be an anecdotal evidence for speech development in these nonverbal participants (Rvachew & Brosseau-Lapré, 2012).

668

8 4.4 Limitations and Clinical Implications

This study has several limitations. First, as with many other studies of autism, a limitation 669 of the current training study is its small sample size, and replication in larger-scale randomized 670 studies will be necessary. Second, more treatment sessions or some other training approaches 671 should be applied to help nonverbal children with ASD improve their speech production skills. 672 Third, considering the substantial heterogeneity of the autism spectrum, there are various types 673 674 of speech disorder in ASD, such as motor speech disorder (dysarthria or apraxia), speech delay, or combination of these (Chenausky et al., 2019). More in-depth investigation of speech therapy 675 in different subtypes would help determine whether MMLI is effective for all children with 676 677 autism or whether it only works well for certain subtypes. Understanding these mechanisms will help tailor the interventions, select the most appropriate personalized treatment, and make 678 679 predictions about prognosis. Fourth, applying the current MMLI training to some other tonal 680 language speakers with ASD would be an important next step.

Taken together, the AMMT-based training program of MMLI, notwithstanding its
limitations, provided an effective training approach in accelerating the rate of speech sound
(especially lexical tone) and word production for Mandarin-speaking children with ASD. The

languages of the world exhibit great diversity. Some estimates suggest that around 60–70% of 684 the world's languages are tonal (Yip, 2002), and more than half of the world's population speak 685 a tone language (Fromkin, 1978). Thus, there is a high demand for MMLI, which could be 686 modified and applied to help some other tone-language-speaking children with autism beyond 687 Mandarin-speaking ones to better acquire the phonological category of lexical tones. With 688 689 respect to practical significance, the current MMLI approach is realized in the smartphone/iPad app, which is easily accessible and has the potential to be utilized remotely in the home 690 691 environment as implemented by a parent or family member. This is important for speech therapy 692 for children with autism from counties with a shortage of speech-language pathologists. Finally, the success of AMMT-based MMLI also lends support to the positive effects of music-supported 693 treatments in individuals with ASD (James et al., 2015; Reschke-Hernández, 2011; Salomon-694 Gimmon & Elefant, 2019; Sharda et al., 2018). 695

696

697 **5** Conclusion

This study compared the efficacy of MMLI, an AMMT-based treatment, with the control 698 therapy in eliciting spoken language for tone-language-speaking children with autism. Relative 699 700 to the control treatment, there was greater improvement in Mandarin-speaking children with ASD after they received MMLI training, in terms of lexical tone, final, and word learning in the 701 702 trained items. Such enhanced training efficacy on lexical tone production persisted at two weeks 703 post-therapy, and even generalized to untrained items that were not practiced. The results hold promise for the efficacy of MMLI to improve speech production in tone-language-speaking 704 705 children with autism. Because the nonverbal and low-verbal children with autism had a very 706 limited repertoire of speech sounds prior to treatment, the acquisition of speech sounds and

words through MMLI is an important gain that provides a foundation for subsequent speech and
language rehabilitation. More importantly, this study offers the first empirical evidence of the
advantages of utilizing musical elements to facilitate lexical tone acquisition in the clinical
population of ASD, which adds a new clinical perspective to our understanding of the close
relationship between music and speech.

712

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994 <u>Tables</u>

Table 1

997 Characteristics of children with ASD and age-matched TD children

	ASD (<i>n</i> = 30)		TD (<i>n</i> = 30)		+	
	М	SD	М	SD	<i>l</i>	p
Age (in months)	67.80	15.06	67.57	14.12	0.56	.578
Language Ability	31.67	26.84	88.07	8.81	-11.81	<.001
Nonverbal IQ	59.53	12.32	105.83	17.34	-13.78	<.001
Working Memory	3.90	4.39	12.23	3.65	-12.00	<.001
% Initials Correct	18.69	16.71	69.74	13.81	-14.23	<.001
% Finals Correct	17.60	15.59	66.31	13.74	-14.05	<.001
% Tones Correct	17.59	15.71	68.38	14.74	-14.24	<.001
% Morphemes Correct	22.68	20.99	73.88	14.73	-12.30	<.001

Table 2

	MMLI (n = 15)		SRT (1	<i>i</i> = 15)		
	М	SD	М	SD		р
Age (in months)	66.13	15.32	69.47	15.14	-0.63	.542
Language Ability	33.07	25.35	30.27	29.08	0.47	.643
Nonverbal IQ	60.53	13.86	58.53	10.97	0.65	.528
Working Memory	3.93	4.38	3.87	4.55	0.05	.962
% Initials Correct	18.49	17.81	18.90	16.17	-0.28	.785
% Finals Correct	17.88	17.05	17.32	14.60	0.22	.830
% Tones Correct	17.90	17.71	17.28	14.05	0.16	.872
% Morphemes Correct	22.44	23.48	22.92	19.02	-0.15	.887

Characteristics of participants with ASD in two treatment groups

Table 3

- 1007 The warm-up stage at the beginning of each treatment session and the five-step structure of an
- 1008 MMLI trial vs. an SRT trial

	MMLI (Experimental Group)	SRT (Control Group)
Warming Up	Musical melodies without lyrics are played, and musical toys such as shaking maracas are introduced to facilitate their movements. Moreover, a rhythmic tapping of the foot is also used in time to music.	Playing checkboards without verbal or with minimally verbal instruction.
Steps	MMLI Trial	SRT Trial
1. Word Introduction	Therapist introduces the target word by showing a word picture (such as "tiger") on the phone/iPad app and then intoning (singing) the word "[lou35 xu214]" by tapping the piano icons 1× per syllable.	Therapist introduces the target word by showing a word picture (such as "tiger") on the phone/iPad screen and then speaking the word "[lou35 xu214]" without finger tapping.
2. Synchronous Production	Therapist produces target with the child. Therapist intones and taps "Let's sing it together" and in synchrony with child "[lo u35 xu214]".	Therapist produces target with the child. Therapist speaks "Let's speak it together" and in synchrony with child "[lqu35 xu214]".
3. Unison with Fading	Therapist and participant begin to intone and tap the target word together, but after the first syllable, the therapist stops while the child continues to intone and tap the next syllable. "[lqu35]".	Therapist and participant begin to speak the target word together, but after the first syllable, the therapist stops while the child continues to produce the next syllable. "[1 gu35]".
4. Immediate Imitation	Therapist firstly intones and taps the target word alone. Afterwards, participant imitates the word, and therapist remains silent. "My turn first: [lou35 xu214]. Now your turn:".	Therapist firstly speaks the target word alone. Afterwards, participant imitates the word, and therapist remains silent. "My turn first: [lqu35 xu214]. Now your turn: ".
5. Independent Production	The child is further encouraged to independently intone and tap the target word once again. ""	The child is further encouraged to independently speak the target word once again. ""

1013 Means (and standard deviations) of % Initials Correct for low-verbal participants by treatment

1014 group (MMLI and SRT) and probe assessment in (a) Trained Items, and (b) Untrained Items.

	Group -	Pretest		Midtest		Immediate Posttest		Delayed Posttest	
		М	SD	М	SD	M	SD	M	SD
(a)	MMLI	23.41	11.83	26.93	11.35	38.48	10.45	36.80	10.98
Trained	SRT	24.03	13.27	27.92	12.90	38.03	11.51	38.34	12.63
(b)	MMLI	21.66	13.95	22.85	17.64	28.17	18.11	27.82	14.97
Untrained	SRT	21.94	14.47	22.45	18.62	28.42	15.18	26.91	12.55

Means (and standard deviations) of % Finals Correct for low-verbal participants by treatment
group (MMLI and SRT) and probe assessment in (a) Trained Items, and (b) Untrained Items.

	Group	Pretest		Midtest		Immediate Posttest		Delayed Posttest	
		М	SD	М	SD	M	SD	M	SD
(a)	MMLI	22.93	12.92	25.54	14.02	37.98	12.61	35.60	12.04
Trained	SRT	22.42	14.28	26.25	13.37	34.21	13.55	33.78	14.02
(b)	MMLI	19.54	14.31	20.36	14.62	27.71	17.58	23.51	15.46
Untrained	SRT	18.03	16.93	18.57	15.18	24.89	14.37	19.08	13.89

Means (and standard deviations) of % Tones Correct for low-verbal participants by treatment
 group (MMLI and SRT) and probe assessment in (a) Trained Items, and (b) Untrained Items.

	Group	Pretest		Midtest		Immediate Posttest		Delayed Posttest	
		М	SD	М	SD	M	SD	M	SD
(a)	MMLI	22.43	9.83	30.32	7.44	45.01	5.47	42.02	5.67
Trained	SRT	21.57	11.54	26.37	10.55	32.85	11.70	33.03	13.42
(b)	MMLI	22.53	15.06	26.03	14.08	34.78	14.95	34.03	14.90
Untrained	SRT	22.42	14.84	22.19	17.45	27.27	15.27	25.57	15.14

1027 Means (and standard deviations) of % Morphemes Correct for low-verbal participants by

treatment group (MMLI and SRT) and probe assessment in (a) Trained Items, and (b) Untrained Items.

	Group	Pretest		Midtest		Immediate Posttest		Delayed Posttest	
		М	SD	М	SD	M	SD	М	SD
(a)	MMLI	28.71	14.03	36.17	8.22	50.87	8.00	47.72	9.18
Trained	SRT	29.05	12.64	35.83	13.20	45.32	13.30	44.35	14.82
(b)	MMLI	24.96	17.97	28.62	13.82	37.62	14.10	34.41	14.73
Untrained	SRT	27.02	15.24	26.04	19.45	34.48	17.95	29.07	17.33





- *Figure 2*. The user interface of the app: (a) the home page of six themes, (b) the interface of one
- 1040 lexical item.



Figure 3. The spectrograms (the upper row) and pitch contours (the bottom row) of the lexical
item "老虎" (tiger) [lou35 xu214] in (a) natural speech sounds, and (b) piano-timbre nonspeech

sounds. The two types of sounds share exactly the same pitch contours with blue curves.

1046



1048 Figure 4. The probe assessments and training procedure for two treatment groups (MMLI and



1050 second-round Midtest, second-round Immediate Posttest, respectively.



1052 *Figure 5.* The production accuracy of initials, finals, tones, and morphemes for low-verbal

1053 participants by treatment group (MMLI and SRT) and probe assessment in the (a) Trained Items,

and (b) Untrained Items. Tests 1–4 represent the Pretest, Midtest, Immediate Posttest, and

- 1055 Delayed Posttest respectively. ***p < .001; **p < .01 after Tukey adjustment for the comparison
- 1056 of MMLI vs. SRT. Error bars: +/- 1 Confidence Interval.