

## **Response to Music-Mediated Intervention in Autistic Children with Limited Spoken Language Ability**

Angela MacDonald-Prégent<sup>1</sup>, Fauzia Saiyed<sup>2</sup>, Krista Hyde<sup>3</sup>, Megha Sharda<sup>3</sup>, Aparna Nadig<sup>1</sup>

1 - School of Communication Sciences and Disorders, McGill University, 2001 Av. McGill

College #8, Montréal, QC, H3A 1G1

2 - Department of Psychology, McGill University, 2001 Av. McGill College #4, Montréal, QC,

H3A 1G1

3 - International Laboratory for Brain, Music and Sound Research (BRAMS), University of Montréal, Pavillon Marie-Victorin/ Local A-108, 90 Vincent-d'Indy Av., Outremont, QC, H2V 2S9

### **Abstract**

*Purpose:* Autistic children with limited spoken language ability (LSLA) often do not respond to traditional interventions, reducing their social inclusion. It is essential to identify effective interventions, and sensitive measures to track their intervention response.

*Methods:* Using data from an RCT comparing music-mediated and play-based interventions, we investigated the impact of spoken language ability on outcomes, and measured response to intervention through natural language sample measures.

*Results:* Children with lower verbal IQ, relative to higher verbal IQ, made some greater gains over the course of music-mediated intervention. Natural language samples were helpful in characterizing communication and tracking change.

*Conclusions:* Music-mediated interventions hold promise as effective interventions for autistic children with LSLA. Natural language samples are robust in characterizing this subgroup.

### **Keywords**

response to intervention, autism, music-mediated intervention, limited verbal ability, minimally verbal, non-verbal

### **Corresponding Author Information**

Correspondence to Angela MacDonald-Prégent using email: [angela.macdonald@mail.mcgill.ca](mailto:angela.macdonald@mail.mcgill.ca)

Spoken language ability is highly variable among people on the autism spectrum<sup>1</sup> (Anderson et al., 2007). Some degree of formal language difficulty is present in approximately half of autistic children (Kjelgaard and Tager-Flusberg, 2001; Pickles et al., 2014) ranging from those who can speak fluently but struggle with grammatical or semantic aspects of formal language to those who are nonspeaking. When measured at preschool age, stronger spoken language in autistic children positively predicts their social-adaptive functioning, socialization, and independent living outcomes at adolescence (Venter et al., 1992) and adulthood (Howlin et al., 2004). Moreover, even as adults, stronger spoken language ability, such as higher vocabulary, continues to predict better vocational and social outcomes (Friedman et al. 2019).

Given the pivotal role of spoken language in outcomes, it is important to understand which subgroups of autistic children benefit most from which interventions (Vivanti et al., 2014). Response to intervention analyses examine how different predictors are impacted by a particular intervention, and which are associated with favorable intervention outcomes. One type of predictor that can be examined are baseline participant characteristics, such as spoken language ability (Trembath and Vivanti 2014; Vivanti et al., 2014). When participating in interventions that target spoken or non-spoken language, children on the autism spectrum who have lower baseline spoken language ability make fewer gains in communication, daily living, social, and motor skills when compared to their autistic peers with higher baseline spoken language (Fossum et al., 2018; Gordon et al., 2011; Itzhak and Zachor, 2011). In order to identify interventions that are appropriate for children with autism who have different levels of spoken language ability (Lombardo et al., 2015;

---

<sup>1</sup> Following recommendations on the use of terminology for autism (Bottema-Beutel et al., 2021, CASDA, 2020), we use identity-first terms such as “autistic person” or neutral terms such as “person on the autism spectrum.” In addition, we use the terms “spoken” or “speaking” rather than “verbal” due to their preferential acceptance by the autistic community.

Lord, Risi and Pickles, 2004; Tager-Flusberg et al., 2009), it is essential to properly define spoken language ability. In this article, we use the term *limited spoken language ability* as an umbrella term to refer to children on the autism spectrum who have significantly less spoken language than their typically developing peers as well as those who are minimally-speaking, and those who are nonspeaking<sup>2</sup>. *Limited spoken language ability* (LSLA) will be operationalized using verbal IQ in Analysis 1, followed by a more comprehensive characterization in Analysis 2.

Different empirically established interventions (i.e., specific manualized interventions, backed by rigorous high quality studies that demonstrate their effectiveness in supporting people on the spectrum) as well as evidence-based practices (i.e., strategies or approaches supported by scientific evidence that shows they support an autistic learner in achieving positive outcomes) have been identified for children on the autism spectrum (Hume et al., 2021; National Autism Center, 2015; Wong et al., 2015). Of direct relevance, *music-mediated interventions* are one of 28 evidence-based practices identified by Hume et al., (2021). Relative to control interventions, music-mediated interventions demonstrate positive gains in children on the autism spectrum with respect to speech, expressive language, social-communication, and parent-child relationships (Bharathi et al., 2019; Chenausky et al., 2016; Geretsegger et al., 2014; Sharda et al., 2018; Thompson et al. 2014). Positive outcomes in social-communication include improvements in initiating, responding, and maintaining social interactions as well as being able to understand another person's perspective. As reviewed by Brancatisano et al., (2020) for neurological disorders more broadly, and by Srinivasan and Bhat, (2013) and Hume et al., (2021) with respect to autism specifically, music-mediated interventions vary in format of implementation and intervention

---

<sup>2</sup> Please see Supplemental Information – Section One for terms and definitions proposed for different segments of LSLA subgroup in the literature.

goals, but share music as a key feature of the intervention delivery. This includes music therapy, which occurs in a therapeutic relationship with a trained music therapist, in addition to the planned use of songs, melodic intonation, and/or rhythm to support the learning or performance of target behaviors and skills in varied contexts (Hume et al., 2021, p. 4024).

Extending work on music-mediated therapy for aphasia, Wan and Schlaug (2010) pioneered the idea that music-mediated interventions could hold promise for children with limited spoken language ability, in particular, those who are nonspeaking. They and others have motivated the use of music-mediated interventions in autism with findings of the neuroplasticity that results from music-making in neurotypical and other populations (see Sihvonen et al., 2017 for a review). With respect to autism, differences observed in neural connectivity between auditory and motor regions can be engaged and potentially improved by music-making, for example listening to someone sing while tapping a drum (Janzen et al., 2018; Srinivasan and Bhatt, 2013; Wan and Schlaug 2010; Wan et al., 2011). Additionally, music and speech processing are subserved by some of the same fronto-temporal circuits which are typically activated when listening to sung, but not spoken, words by children on the autism spectrum (Sharda et al., 2015), suggesting that music may be an alternative route that could be capitalized on during intervention.

Prior studies testing the effectiveness of music-mediated therapies for children with limited spoken language included minimally or nonspeaking children and employed interventions such as Auditory-Motor Mapping Training which involve imitating sung words in conjunction with motor activity (e.g., tapping on a drum), with the outcome measure of children's ability to speak the taught words (Chenausky et al., 2016; Wan et al., 2011; Sandiford et al., 2013). The music-mediated approach we investigate here is quite different in format and target, as implemented in our prior randomized controlled trial (Sharda et al., 2018). It consisted of sessions where a music

therapist engaged an autistic child in shared music-making activities that targeted communication, social reciprocity, sensorimotor integration, and emotion regulation. A loose structure was followed across participants, while adapting interaction to the individual child's needs. This type of approach has been termed improvisational music therapy (see Geretsegger et al., 2014; Srinivasan and Bhat, 2013). In this randomized control trial (RCT), we demonstrated a significant increase in social-communication in autistic children who took part in this music-mediated intervention (MI), as compared to those who participated in a control play-based intervention (nonMI) which shared the same intensity and intervention targets. Moreover, we found increased functional connectivity between auditory and motor brain regions in children in the MI group, which was related to behavioral improvements in their communication skills (Sharda et al., 2018).

In an RCT of improvisational music therapy by Crawford et al. (2017), the authors explored relationships between baseline characteristics (e.g., gender, nonspeaking status) and intervention outcomes in 304 children. They found that a larger proportion of nonspeaking children made improvements on the social affect scale of the Autism Diagnostic Observation Schedule (Lord et al., 2001;  $RR^3$ : 1.45), compared to nonspeaking participants in the intervention-as-usual condition. The intervention-related difference for nonspeaking participants was greater than the improvement observed in the full sample on the same metric ( $RR$ : 1.25, Crawford et al., 2017, Figure 3). Sharda et al., (2019) highlighted these findings, and echoing Wan and Schlaug (2010), hypothesized that MIs, including those broader in approach than those teaching specific speech sounds, might be particularly beneficial for autistic children who have limited spoken language. We examine this

---

<sup>3</sup> Risk ratio ( $RR$ ) is defined as the probability of a condition (in this case, type of intervention), affecting the outcome (e.g., improvements on the ADOS social affect scale). Here,  $RR = 1$  indicates the intervention conditions have equal effect on the outcome,  $RR > 1$  indicates an increased effect of improvisational music therapy on the outcome,  $RR < 1$  indicates an increased effect of intervention-as-usual on the outcome.

hypothesis, using data from our prior RCT (Sharda et al., 2018) to conduct post-hoc analyses. In Analysis 1, we examine how baseline spoken language ability impacts response to MI with a sample of school-age autistic children. Since traditional interventions are sometimes not optimal for autistic children with limited spoken language ability, we hypothesize that MI, an intervention that is less language-mediated but still targets communication skills, will be more effective for them.

A second goal of this study is careful characterization of communication in children with limited spoken language and identifying appropriate outcome measures for them. Children with this language profile are often classified using only one domain of spoken language (Koegel et al., 2020). As children on the autism spectrum often present with uneven or asymmetrical difficulties across domains of language such as phonology, vocabulary, syntax and pragmatics (Tager-Flusberg et al., 2009), their spoken language ability should be characterized using several language domains. Another challenge that exists for this subgroup is the ability to sensitively measure and track intervention outcomes (Kasari et al., 2013; Koegel et al., 2019; Trembath et al., 2019). Children with limited spoken language ability are often unable to complete common standardized tests of language due to a lack of prerequisite skills (Koegel et al., 2020; Trembath et al., 2019) or floor effects (Barokava et al., 2021; Barokava and Tager-Flusberg, 2018). For example, formal tests of spoken language ability require the child to follow increasingly complex instructions in an assessment context which often does not reflect a child's ability to spontaneously use spoken communication in real life. For these reasons, it is important to identify alternative tools that are better able to track spoken language and social communication as intervention progresses.

Kasari et al. (2013) called attention to the natural language sample (NLS) as a viable tool to access the functional spoken communication skills of minimally speaking autistic children. An

NLS is an objective yet flexible method of providing detailed information on spoken language and non-spoken communication in a natural setting that does not require any pre-requisite language skills, and is not subject to floor effects (Barokava et al., 2021; Barokava and Tager-Flusberg, 2018), and is thus an excellent choice for people with limited spoken language ability (Barokava et al., 2020, 2021; Chiang, 2009; Kasari et al. 2014; Paul et al., 2013). Barokava et al., (2021) developed and tested a language sample protocol, Eliciting Language Samples for Analysis (ELSA), obtained through eight play, narrative, and conversation activities (20-25 minutes in duration). The ELSA protocol and scoring procedures can be used reliably with autistic children and youth who have a range of spoken language ability, including those who have limited spoken language ability (Barokava et al., 2021).

As described below, our findings from Analysis 1 indicate that autistic school-age children with limited spoken language (measured by verbal IQ) do respond better to MI than a control intervention. In Analysis 2 we go one step further by (1) carefully characterizing limited spoken language ability in the Sharda et al., (2018) sample, using a combination of spoken language measures (including NLS measures) and (2) tracking change over the course of intervention using these measures. While conducting an independent NLS session using the ELSA NLS standardized elicitation protocol would be ideal, it is not always feasible. For example, when conducting assessment remotely where elicitation materials and trained personnel are not on-site with the participant, or in our case when conducting a post-hoc analysis using available video data. Our interest was to compare different measures of varying resource-intensiveness, rather than to employ one measure comprehensively across all sessions. In addition to using some of the spoken language measures used by ELSA (frequency of utterances, mean length of utterance, and number of different words) (Barakova et al., 2020 and 2021), we included a more global measure of



communication: frequency of intentional communication acts (ICA; Yoder et al., 2015). ICA accounts for both intentional spoken and non-spoken communication, allowing for use with children who are non speaking. Though exploratory in nature, we hypothesize that the NLS procedure will more clearly and comprehensively characterize communication in this subgroup, and that NLS measures will be sensitive in tracking change over the course of intervention.

### **Methods: Analysis 1**

#### *Participants*

Our prior RCT (Sharda et al., 2018) was conducted with 51 six- to twelve-year-old children on the autism spectrum who participated in a MI or nonMI. All participants were reported to have no recent history of individual music therapy or music lessons and did not attend group music therapy at school. The current analyses examine whether spoken language is a predictor of response to interventions that were delivered in English. Therefore, we retained 47 children who had English as a primary language, excluding four children from the original RCT who did not meet this criterion. Verbal IQ was the spoken language predictor selected for Analysis 1 (see Language Measure Selection – Predictor Variable section below) but was found to be non-equivalent between the MI and nonMI groups at baseline (i.e.,  $d = 0.39$ , variance ratio = 1.12). To match the groups on verbal IQ based on guidelines provided by Kover and Atwood (2013), two participants with the highest verbal IQ from MI and three participants with the lowest verbal IQ from nonMI were removed. This resulted in a final sample of 42 participants for Analysis 1 whose participant characteristics and outcome measures at baseline are shown in Table 1.

All 42 children met DSM-IV criteria (Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition criteria) for Autism Spectrum Disorder. Verbal IQ was measured using the Wechsler Abbreviated Subtest of Intelligence – 2<sup>nd</sup> Edition (WASI-II; Wechsler, 1999) and

ranged from 55 to 130, with eight participants with a verbal IQ above the average range (that is, above a standard score of 115 and the 84th percentile), twelve with a verbal IQ in the normal range (between 85-115, or the 16th to 84th percentiles), and 21 with a verbal IQ below the average range (below 85 or the 16th percentile). Verbal IQ information was unavailable for one participant who had very limited spoken language skills, thus prohibiting administration of this measure. Importantly, both MI and nonMI groups are equivalent in baseline verbal IQ (i.e.,  $d < .01$ , variance ratio = 1.22).

### *Intervention*

Of the 42 participants included in Analysis 1, 21 were randomly assigned to receive music-mediated intervention (MI) and 21 to a play-based control intervention (nonMI) in the original RCT (Sharda et al., 2018). MI consisted of music-mediated activities incorporating musical instruments, using a child-centric approach (Bradt, 2012; Guerrero et al., 2014; Mössler et al., 2019; Nordoff and Robbins, 2007) whereas, the active control, nonMI, used play-activities and materials to support and engage the child. Both interventions were delivered in English and matched on dosage (i.e., 45 minutes weekly sessions for 8-12 weeks), format (i.e., individual sessions), and target areas (i.e., communication, social reciprocity and turn taking, sensorimotor integration, and emotional regulation). MI and nonMI were delivered by the same accredited music therapist in the same music center using the same session structure: (1) opening: hello song and greeting, (2) choosing the order of four pre-selected activities using a schedule board, (3) completion of four activities, (4) closure: goodbye song and farewell. See supplemental information, section two (Figure and Table S1) and Sharda et al., (2018) for more details on the implementation of the intervention.

### *Outcome Variables*

We examined the impact of intervention group (MI or nonMI) and baseline spoken language ability on two outcomes reflecting key areas of difficulties in autism (DSM-V, American Psychiatric Association, 2013): a) level of coordinated joint engagement, or attention to and engagement with another person while involved in joint activities (Adamson et al., 2004) and b) social communication ability and autism-related behaviors. The measures we chose for these domains are detailed and appropriate to characterize behavior in children with less spoken language; thus, they are potentially sensitive outcome measures for this population.

Joint engagement, recently highlighted as an active ingredient of music-mediated interventions (Latif et al., 2021), can be defined as engagement involving two individuals and an event whereby the individuals jointly attend to each other and participate in a shared event (Girolametto et al., 1994). We adapted Adamson et al. (2004)'s joint engagement coding scheme, previously shown to be sensitive to intervention effects (Kasari et al., 2010) in a school-age autistic population, to our MI and nonMI intervention activities. Our coding scheme (Latif et al., 2021) consists of four categories of joint engagement: *coordinated joint*, *supported joint*, *object*, and *other*. Here we focus on the highest level of joint engagement, coordinated joint engagement, exclusively and examine potential interactions with verbal IQ. Coordinated joint engagement states were coded when the child and the therapist were simultaneously engaged in the same activity and the child initiated spoken or gestural communication directed to the therapist. Videos from the intervention sessions (first and last) were coded by independent raters blind to timepoint, intervention and hypotheses. Reliability for the coordinated joint code ( $ICC = 0.70$ ,  $95\% CI = 0.57-0.79$ ) was moderate, following guidelines by Koo and Li (2016), though other codes reached a good level of reliability (Latif et al., 2021). The change score in percent time spent in a coordinated joint engagement state between the last and first intervention session (i.e., last

intervention session - first intervention session/ first intervention session + last intervention session \* 100) was used as the outcome variable for this analysis.

Social communication and autism-related behaviour outcomes were obtained using a parent report questionnaire, the Children's Communication Checklist – 2<sup>nd</sup> edition (CCC-2, Bishop, 2003). The CCC-2's general composite score was reported as a primary outcome in Sharda et al., 2018. Here, we examine another combination of scales in the CCC-2, the pragmatic-autism composite score<sup>4</sup> (CCC-Pragaut), an unstandardized composite score obtained by summing six CCC-2 subscales that address the ability to appropriately initiate, the use of stereotyped language, pragmatic communication, non-verbal communication, social interaction, social awareness, and the presence or absence of autism-type behaviours such as restricted and repetitive interests. Unlike the general composite score, the CCC-Pragaut does not include scales that focus on structural language such as semantics, syntax, speech, and coherence. The CCC-Pragaut was chosen due to its breadth of constructs related to social-communication and autism-type behaviours that could be used to evaluate change in daily life. The CCC-Pragaut has successfully identified social communication difficulties (Leonard et al., 2011; Timler, 2014) in various clinical populations (i.e., children with attention deficit/hyperactivity disorder) in comparison to neurotypical children.

#### *Language Measure Selection – Predictor Variable*

For this post-hoc analysis, spoken language ability was the predictor of interest. Three measures, with no significant differences between intervention groups at baseline, were identified: i) the

---

<sup>4</sup> Although this composite score has been termed “Pragmatic-autism” in the prior literature, we consider it to be a measure of broader social communication skills (see subscales included) rather than of language pragmatics alone (See Norbury (2014) pp.209-210 for a discussion of this distinction).

sentence repetition subtest from the Clinical Evaluation of Language Fundamentals – 4<sup>th</sup> edition (CELF-4SR; Semel et al., 2004), ii) the Peabody Picture Vocabulary Test, a receptive vocabulary test (PPVT-4; Dunn and Dunn; 2007), and iii) the verbal IQ subtest score from the WASI-II (Wechsler, 1999). The WASI-II verbal IQ subtest score was chosen to be the spoken language ability predictor variable for this analysis as 1) it focused on spoken language ability unlike the PPVT-4 and 2) in comparison to the CELF-4-SR, provided more robust information on spoken language (i.e., assessed spoken language through labelling items and identifying similarities as opposed to simply repeating a sentence). Please see supplemental information, section three for a further description of the WASI-II.

Finally, the intervention group was included as an independent variable based on findings from Sharda et al. (2018) where gains in social-communication were significantly higher for participants in the MI group. If MI is indeed more effective for children with limited or lower spoken language ability than a nonMI control intervention, we would expect to observe a significant interaction between spoken language ability and intervention group.

#### *Variable Transformation and Statistical Analysis*

The data set had a total of 13 missing data points in which 12 were imputed. Please see supplemental information, section four for details on the missing data and imputation techniques. Two fixed effects multiple linear regression models with effects of intervention group (MI vs. nonMI), spoken language ability (verbal IQ score), and their interaction were estimated for each outcome measure, coordinated joint engagement and CCC-Pragaut. Linear regression models were carried out in R version 3.6.2 (R Core Team, 2013) using the `lm` function in the tidyverse package. To help facilitate interpretation of the resulting models, the intervention group variable was relevelled to make nonMI the reference level, the CCC-Pragaut was transformed from a raw to z-

score as it was unstandardized, and verbal IQ was centered. Model diagnostic plots for both the coordinated joint and CCC-Pragmat models were examined and assumptions for linearity and normality were generally met.

## **Methods: Analysis 2**

### *Participants*

The goal of Analysis 2 was to characterize and track the outcomes of autistic children with limited spoken language abilities, irrespective of intervention group membership. Therefore, we conducted two rounds of screening, starting with all 47 children who had English as a primary language. Please see supplemental information, section five for details on the screening procedure. Of these, 19 participants were identified as potentially having limited spoken language for more detailed characterization using our multi-criteria definition described below.

### *Multi-Criteria Definition of Limited Spoken Language*

A multi-criteria definition of limited spoken language ability (LSLA) was created to capture ability in three spoken language areas: 1) spoken language imitation ability, 2) lexical diversity, and 3) syntactic complexity. First, performance on the CELF-4-SR subtest, a standardized measure of spoken language repetition, was also used. Children needed to receive a scaled score of 4 or less, indicative of below average performance. Four participants who obtained a scaled score of 6 or above for the CELF-4-SR were excluded, as they demonstrated an average or above average ability to repeat sentences and thus indicating that their spoken language ability was not limited. From there, 15 children were analyzed using the natural language sample transcription procedure which provide the second and third criteria for the definition of LSLA. Second, children needed to have a total number of DRW (different root words) that was two standard deviations or greater below the mean provided by Leedholm and Miller (1994). A root word is defined as a spontaneous,

intelligible, and unique root word present in an NLS (Miller et al., 2016). For example, the words *go* and *going* would be counted as 1 root word. Finally, a mean length of utterance in words (MLUw) of three words or below was required. MLUw is the average number of words in an utterance. This cut-off was chosen as it closely resembles the minimally speaking or low spoken skill inclusion criteria used in Pecukonis et al. (2019). This is based on the administration criteria of the ADOS-2 module one (Lord et al., 2012), and is the currently most used criterion to define minimally speaking participants (Bal et al., 2016). The MLUw and DRW data were collected using an NLS from each participant's first intervention session (described in detail below). All three criteria were required to capture difficulties in both length and variation in spoken language and provide a comprehensive multi-criteria definition of limited spoken language. One participant met the CELF-4-SR and DRW criteria but had an MLUw of 4.52 and therefore was excluded from Analysis 2 resulting in a final sample of 14 participants, 6 in the MI group and 8 in the nonMI group.

#### *Natural Language Sample Coding Procedures*

Videos from the first (timepoint 1) and last (timepoint 2) session in our prior RCT (Sharda et al., 2018) were used for natural language sampling. Timepoint 2 was 7 to 11 weeks later where eight of the fourteen participants completed all twelve sessions, two completed 11/12 sessions, two completed 10/12 sessions, and 2 completed 9/12 sessions. As mentioned, MLUw and DRW at timepoint 1 were used as a part of the multi-criteria definition of *limited spoken language ability*. The first 15 minutes<sup>5</sup> of spoken language-based interactions were transcribed and coded for MLUw and DRW. In addition, the number of spontaneous and intelligible spoken utterances, referred to

---

<sup>5</sup> Note that the first 15 minutes for children in the MI group were collected in between musical activities resulting in a longer total time. Musical activities were not coded as they did not involve any spoken interactions.

as non-imitative spoken communication acts (NISCA) were also tabulated (Yoder and Stone, 2006). Finally, non-spoken intentional communication was coded using the definition of intentional communication act (ICA): a spoken or nonspoken act that serves to intentionally communicate a message (Yoder et al., 2015). For example, an ICA can be conventional or non-conventional gestures, sign language, intentful non-word vocalizations, or imitative symbols that are coordinated towards a communication partner.

NLS video transcription and coding was evenly split between the first author and a second coder who was blind to condition and timepoint and had not been involved in screening. The second coder was introduced to the procedures using an in-house NLS manual. Transcription of spoken language followed the Systematic Analysis of Language Transcripts (SALT) procedures outlined by Miller et al. (2016) but was adapted to include instructions on how to transcribe play and music-mediated intervention videos. Following this, the first author and second coder jointly coded three training videos. Coding discrepancies were discussed, and adjustments were made to the manual. Once training was completed, they transcribed and coded 14 videos each. The total dataset included twenty-eight 15-minute NLS videos (e.g., 14 participants x 2 timepoints).

### *Inter-Rater Reliability*

The first author and blind coder double transcribed and double coded 19 of the 28 videos, representing 68 percent of the dataset. Intraclass coefficients based on a single rater, agreement, two-way random effects model were calculated using the IRR package in R (version 3.6.2) for each NLS variable. Excellent reliability (e.g., values 0.90 or higher, Koo and Li, 2016) was obtained for all dependent variables: ICA (ICC = 0.98, 95% CI = 0.95, 0.99), NISCA (ICC = 0.99, 95% CI = 0.97, 0.99), DRW (ICC = 0.97, 95% CI = 0.91, 0.99), MLUw (ICC = 0.99, 95% CI = 0.97, 0.99).



### *Statistical Analysis*

Correlations using R version 3.6.3 between NLS variables were obtained to determine the level of collinearity in order to assess the NLS procedures used for characterization. All four NLS variables: MLUw, DRW, ICA, and NISCA, were found to have moderate to strong correlations (see Table 2), demonstrating that all four variables are highly associated. Due to the exploratory nature and small sample size, a full convergent validity analysis was not conducted.

### **Results: Analysis 1**

As shown in Table 3, a significant interaction was found between verbal IQ and intervention group ( $\beta = -0.52$ ,  $t = -2.28$ ,  $p = 0.029$ ) for the Coordinated Joint Engagement model. As illustrated in Figure 1, for children receiving MI, those with lower verbal IQ scores experienced a greater increase in percent time spent in Coordinated Joint Engagement. No significant main effects of verbal IQ or intervention group were found.

For the CCC-Pragmat model, a significant main effect for intervention group ( $\beta = 0.81$ ,  $t = 2.74$ ,  $p = 0.009$ ) shown in Table 4 and illustrated in Figure 2, shows more improvement in the CCC-Pragmat scores in the MI group in comparison to children in the nonMI group, demonstrating an overall improvement in social communication and autism-like behaviours in the MI participant's contexts outside of intervention as reported by their parents. No significant effects were detected for the remaining variables (i.e., verbal IQ, or the interaction between intervention group and verbal IQ).

These two findings indicate improvements in key targets of intervention during the session (i.e., joint engagement coded from session video) as well as generalized improvement to daily life (i.e., parent report of social communication and autism-like behaviours), noting more improvement

in joint engagement for children with lower verbal IQ scores in comparison to those with higher verbal IQ scores.

### **Results: Analysis 2**

Fourteen participants met the multi-criteria definition of LSLA at timepoint 1, 6 from the MI group (LSLA-MI) and 8 from the nonMI group (LSLA-nonMI). Table 5 presents demographic and NLS measures for this subsample at baseline. Figure 3 presents change from timepoint 1 to timepoint 2 in each of the four NLS measures: DRW, MLUw, NISCA, and ICA for this subsample.

As mentioned in the introduction, children with LSLA perform near floor levels on many standardized language measures, rendering these measures incapable of tracking change over the course of intervention. For instance, on the formal language-based measures employed at baseline in the RCT for this subsample, 6 of 14 children were at floor on the CELF-SR (i.e., sentence repetition subtest from the Clinical Evaluation of Language Fundamentals – 4<sup>th</sup> edition), and 12 of 14 were at floor on the WASI-II (i.e., Wechsler Abbreviated Subtest of Intelligence – 2<sup>nd</sup> Edition). In contrast, there are minimal floor effects for the NLS measures. Through visual inspection of Figure 3, change from timepoint 1 to timepoint 2 can be seen for many participants, and this is the case for all four variables. In comparison to DRW and MLUw (Figures 3a and 3b), greater increases in NISCA and ICA (Figures 3c and 3d) can be seen for some participants. This could be in part because NISCA and ICA track the total quantity of spoken and in the case of ICA, spoken and non-spoken communicative bids whereas DRW and MLUw track linguistic complexity (i.e., diversity and length) of spoken communicative bids.

### **Discussion: Analysis 1**

Using data from Sharda et al. (2018) we explored response to MI in autistic children, with a focus on differences in spoken language ability as a potential predictor, given the proposal that

MI may, in particular, benefit children with limited spoken language ability (Chenausky et al., 2016; Sharda et al., 2019; Wan and Schlaug 2010; Wan et al., 2011,). We observed an interaction between verbal IQ and intervention group. Specifically, children with lower verbal IQ scores receiving MI had a significantly greater increase in coordinated joint engagement with the therapist over the course of intervention, compared to children with higher verbal IQ scores who also underwent MI. The reliability of our coordinated joint engagement measure was moderate ( $ICC = .70$ ), similar to the value reported for this code in the original joint engagement coding scheme for toddler-parent interactions (Adamson et al., 2004, p. 1178). This finding complements a finding from another post-hoc analysis (Crawford et al., 2017) where a larger portion of nonspeaking children on the autism spectrum in their MI group made gains in social affect than those in their control group.

Why did we not observe similar gains in joint engagement in children receiving MI who had stronger spoken language ability? We speculate that the children with stronger spoken language ability were able to engage in the activities of the music intervention, independently from the therapist. The music-mediated intervention implemented in Sharda et al. (2018) used a structured format where the focus was exclusively on musical interaction. This may have left little room for autistic children who have stronger spoken language skills to incorporate their own play interests or to express themselves through speech within the musical set up, which we sometimes observed. This could have contributed to less coordinated joint engagement over the course of the MI intervention (see data point in the bottom right corner of Figure 1).

We hypothesize that the play-based activities in the nonMI condition were better a medium for engagement for children with stronger spoken language skills as they were more open-ended (e.g., playdoh, lego, finger puppets), allowing the child to voice their specific play ideas and to

have them mirrored or followed through by the therapist (e.g., child makes a playdoh cat and asks the therapist to make food for the cat) thus potentially leading to more coordinated joint engagement over time (e.g., both engaging in the playdoh activity together). Thompson and Elefant (2019) discuss the need to adapt MI implementation for autistic children who have strong spoken language ability and may engage with music in different ways and may also prefer spoken play activities. They recommend allowing these children to carry out their alternative play activity while the therapist layers in musical components (e.g., the child acts out a play scheme while the therapist plays music to go along with the play scheme) rather than trying to get the child to actively participate in the music component. Though the child is more of a passive participant to the musical experience, they argue that MI can still be used to improve key social and communicative outcomes.

Second, with respect to the outcome of parent-reported social communication and autism-related behaviours, the entire MI group experienced a greater increase in skills than did children who underwent the control play-based nonMI intervention; there were no differences based on spoken language ability. This parallels our prior finding in Sharda et al. (2018), where we found a significant improvement in the MI group compared to the nonMI group on the CCC-2 general composite score, a measure of social-communication that also includes items related to structural language, relative to the CCC-Pragmat score investigated here. This suggests that music-mediated interventions may hold benefits in targeting core areas of difficulties in autism such as social-communication and restricted and repetitive interests, regardless of their spoken ability. This falls in line with the previous MI studies that have reported gains in similar areas (Bharathi et al., 2018, Geretsegger et al., 2014, Sharda et al., 2018, Thompson et al. 2014). Coordinated engagement was measured by blinded coders from intervention session videos, whereas CCC-Pragmat data was

based on parent-report of behaviour observed in daily life and was broader in scope, covering social interaction, non-spoken communication, pragmatics, and autism-type behaviours. On the one hand, coordinated engagement coding in the lab may be a more detailed and sensitive measure for capturing response to short term interventions, on the other hand, CCC-Pragmat reflects behaviours parents observe in generalized settings. Future research using multiple measures of each construct are needed to clarify the difference observed here.

Overall, children with limited spoken language ability, undergoing MI, making more gains in joint engagement than children with stronger spoken language, is an important finding as previous response to intervention research has shown that autistic children who have limited spoken language often do not make as many gains in social-adaptive skills in comparison to their peers with stronger spoken language (Fossum et al., 2018; Gordon et al., 2011; Itzhak and Zachor, 2011). In particular, nonspeaking and minimally speaking autistic children are an understudied subgroup (Tager-Flusberg and Kasari, 2013) and do not have many effective evidenced-based intervention options (Brignell et al., 2018; Koegel et al., 2019; Rose et al., 2016). Therefore, music-mediated interventions represent an intervention where improvement in core areas of difficulties in autism are possible for this subgroup of autistic children (Chenauksy et al., 2016; Crawford et al., 2017; Wan and Schlaug 2010; Wan et al. 2011). However, future research is needed to replicate this finding and continue investigating interventions that are effective for autistic children who have limited spoken language ability.

### **Discussion: Analysis 2**

Of the 47 children with English as a primary language from the larger RCT sample, 14 met our multi-criteria definition of Limited Spoken Language Ability (LSLA) for further analysis. Although this intensive characterization of spoken and non-spoken communication is not practical

to implement in all contexts, a robust description is required as this sub-population has discrepant ability across language domains (Tager-Flusberg et al., 2009). Our NLS variables sampled multiple domains including syntactic complexity, lexical diversity, and quantity of spoken communication and also the global measure of intentional communication acts which can be used with children who are nonspeaking as it measures non-spoken communication. All NLS variables exhibited moderate to high inter-correlations. Therefore, only one measure may need to be coded as the primary intervention outcome, thereby decreasing the time demand that this procedure presents. However, it is recommended that this be confirmed using a larger sample.

This exploratory analysis further adds support to the use of NLS to track outcomes in intervention. In comparison to the sentence repetition task from the CELF-SR and the spoken language verbal intelligence subtests from the WASI-II, DRW, MLUw, and NISCA presented with minimal floor effects and only one participants' MLUw was not coded as they did produce enough utterances to calculate it. This demonstrates NLS variables are sensitive enough to track change across intervention in autistic children with LSLA.

### **General Discussion**

Here we examined response to intervention in autistic children who have limited spoken language ability, using data from a prior RCT (Sharda et al., 2018) comparing music-mediated (MI) and play-based (nonMI) interventions for school age children on the autism spectrum. The type of MI employed in this RCT involved improvised shared music-making between therapist and child and did not focus on spoken language specifically, but rather targeted the broader intervention goals of communication, social reciprocity, sensorimotor integration, and emotional regulation. In Analysis 1, we examined how baseline spoken language ability, measured by verbal

IQ, and type of intervention impacted outcomes in the key areas of 1) joint engagement with the therapist during the intervention and 2) parent-reported social communication skills and autism-related behaviours. First, we found that children with limited spoken language (i.e., lower verbal IQ) who underwent music-mediated intervention had a significantly greater increase in percent time spent in coordinated joint engagement in comparison to those with stronger spoken language ability undergoing the same intervention. While this finding could reflect that music-mediated interventions are potentially well-suited for children with limited spoken language ability (Sharda et al., 2019; Wan and Schlaug, 2010) and less-suited for children with stronger spoken language ability, it must be interpreted with caution given the small sample size and because it reflects a particular implementation of music-mediated intervention which may not have been ideally suited for children with stronger spoken language skills (Thompson and Elefant, 2019), as mentioned in Discussion: Analysis 1. In Latif et al., (2021) we found that both joint engagement, on one hand, and the involvement of movement, on the other, are *active ingredients* or processes that give rise to positive outcomes of music-mediated interventions for children across the autism spectrum, and discuss the evidence supporting these factors at length. Further research is needed to investigate which specific aspects of music-mediated interventions are the most potent active ingredients for children with limited spoken language ability. We suggest, as previously proposed by other researchers (Janzen et al., 2018; Srinivasan and Bhatt, 2013; Wan and Schlaug 2010; Wan et al., 2011) that hearing music while engaging in the movement involved in music-making may be critical to engaging and improving auditory-motor region brain connectivity, and in turn language and communication. This could be implemented, as seen in Crawford et al. (2017), Kim et al. (2008), and Sharda et al. (2018), in a dyadic intervention format involving moving with a therapist

in joint music-making, where the therapist flexibly adapts interaction to the child by imitating, reinforcing or complementing his/her music making, allowing for moments of synchronization.

Second, we observed that all children in the music-mediated intervention group, with a range of spoken language ability, made significantly greater improvements in social communication skills and autism related behaviours than those who participated in play-based intervention. This adds to the literature demonstrating that music-mediated interventions can improve social-communication, a core area of difficulty in autism. However, this outcome measure was based on parent report and may be biased as parents were aware of the intervention group their child participated in.

Overall, these findings partially support the hypothesis that autistic children who have limited spoken language may respond better to music-mediated interventions than autistic children who had stronger spoken language ability by showing that these children became more engaged with their therapist and activities during intervention. It is possible that active participation in musical activities created the optimum circumstances for children with limited spoken language ability to engage.

In exploratory Analysis 2, going beyond verbal IQ, to appropriately characterize and measure change in autistic children who have limited spoken language we developed a comprehensive definition using a combination of measures including those derived from natural language samples across multiple language domains. Natural language sample measures were used to track change from first to last intervention session in this small but well-defined sub-sample.

Unlike traditional standardized measures such as the CELF-SR and the WASI-II verbal IQ subtests, NLS measures such as DRW, MLUw, NISCA, and ICA are more sensitive at detecting change in autistic children with LSLA as these measures are less subject to floor effects and can



be used with children on the autism spectrum with a variety of spoken language ability including those with limited abilities. Future research, using larger evenly matched groups in terms of spoken language ability, should examine the potential impact a non-language focused intervention, such as a music-mediated intervention, has on spoken language in comparison to a language-focused intervention for children with limited spoken language ability.

### *Limitations*

Though the results of Analysis 1 are promising, as it was a post-hoc analysis, leading to decreased internal validity, 1 - this limits what can be concluded regarding the impact that spoken language ability has on joint engagement in the context of music-mediated intervention and 2 -as the original Sharda et al. (2018) RCT was not conceived with the purpose of recruiting autistic children who had limited spoken language ability, this led to a very small sub-sample in Analysis 2. Further replication of this finding is needed.

Given that parents in the RCT were aware of their child's group assignment, future research should include a direct comprehensive measure of communication that is not subject to potential parental bias, in addition to parent report on the CCC-2. In terms of comprehensive measures of communication, children with LSLA often demonstrate floor effects on standardized measures, so using a natural language sample ( as was done in Analysis 2) is recommended. However, it should be noted that our natural language samples came from available video data of intervention sessions. It would be ideal to employ an independent, standardized elicitation protocol at pre, mid and post intervention, such as the. Eliciting Language Samples for Analysis (Barokova et al., 2021). . Future work should explore response to music-mediated versus comparison interventions in much larger samples of autistic children who have limited spoken language ability.

### *Conclusion*

In Analysis 1, school-age children on the autism spectrum undergoing music-mediated intervention who had limited spoken language (as measured by verbal IQ) made the most improvement in joint engagement. This is striking because children with limited spoken language often have poorer response to language and communication-based interventions (Fossum et al., 2018; Gordon et al., 2011; Itzhak and Zachor, 2011). Importantly, in addition to music-mediated interventions that teach specific spoken words using melody (Chenausky et al., 2016; Wan and Schlaug, 2010), music-mediated interventions with broader intervention goals such as the one implemented in Sharda et al., (2018) are effective for autistic children who have limited spoken language.

Findings from Analysis 2 demonstrated that measures taken from natural language samples are helpful in characterizing the spoken and non-spoken communicative abilities of autistic children with limited spoken language abilities as traditional measures are often not sensitive enough.

## References

- Adamson, L. B., Bakeman, R., and Deckner, D. F. (2004). The development of symbol-infused joint engagement. *Child development*, 75(4), 1171-1187. <https://doi.org/10.1111/j.1467-8624.2004.00732.x>
- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5®)*. American Psychiatric Pub.
- Anderson, D. K., Lord, C., Risi, S., DiLavore, P. S., Shulman, C., Thurm, A., Welsh, K., and Pickles, A. (2007). Patterns of growth in verbal ability among children with autism spectrum disorder. *Journal of consulting and clinical psychology*, 75(4), 594-604. <https://doi.org/10.1037/0022-006X.75.4.594>
- Bal, V. H., Katz, T., Bishop, S. L., and Krasileva, K. (2016). Understanding definitions of minimally verbal across instruments: Evidence for subgroups within minimally verbal children and adolescents with autism spectrum disorder. *Journal of child psychology and psychiatry*, 57(12), 1424-1433. <https://doi.org/10.1111/jcpp.12609>
- Barokova, M., and Tager-Flusberg, H. (2018). Commentary: Measuring language change through natural language samples. *Journal of autism and developmental disorders*, 50(7), 2287-2306. <https://doi.org/10.1007/s10803-018-3628-4>
- Barokova, M. D., Hassan, S., Lee, C., Xu, M., and Tager-Flusberg, H. (2020). A comparison of natural language samples collected from minimally and low-verbal children and adolescents with autism by parents and examiners. *Journal of speech, language, and hearing research*, 63(12), 4018–4028. [https://doi.org/10.1044/2020\\_JSLHR-20-00343](https://doi.org/10.1044/2020_JSLHR-20-00343)
- Barokova, M. D., La Valle, C., Hassan, S., Lee, C., Xu, M., McKechnie, R., Johnston, E., Krol, M.A., Leano, J., and Tager-Flusberg, H. (2021). Eliciting Language Samples for Analysis

- (ELSA): A New Protocol for Assessing Expressive Language and Communication in Autism. *Autism research*, 14(1), 112–126. <https://doi.org/10.1002/aur.2380>
- Bharathi, G., Venugopal, A., and Vellingiri, B. (2019). Music therapy as a therapeutic tool in improving the social skills of autistic children. *The Egyptian journal of neurology, psychiatry and neurosurgery*, 55(1), 1-6. <https://doi.org/10.1186/s41983-019-0091-x>
- Bishop, D. V. (2003). The Children's Communication Checklist: CCC-2. *Harcourt Assessment*.
- Bottema-Beutel, K., Kapp, S. K., Lester, J. N., Sasson, N. J., and Hand, B. N. (2021). Avoiding ableist language: Suggestions for autism researchers. *Autism in adulthood*. 3(1), 18-29. <https://doi.org/10.1089/aut.2020.0014>
- Bradt, J. (2012). Randomized controlled trials in music therapy: guidelines for design and implementation. *Journal of music therapy*, 49, 120-149. <https://doi.org/10.1093/jmt/49.2.120>
- Brancatisano, O., Baird, A., and Thompson, W. F. (2020). Why is music therapeutic for neurological disorders? The Therapeutic Music Capacities Model. *Neuroscience and biobehavioral reviews*, 112, 600-615. <https://doi.org/10.1016/j.neubiorev.2020.02.008>
- Brignell, A., Chenausky, K. V., Song, H., Zhu, J., Suo, C., and Morgan, A. T. (2018). Communication interventions for autism spectrum disorder in minimally verbal children. *Cochrane database of systematic reviews*, (11). <https://doi.org/10.1002/14651858.CD012324.pub2>
- Canadian Autism Spectrum Disorder Alliance. (June 2020). CASDA-ACTSA Language Guide [PDF file]. Retrieved from <https://www.casda.ca/wp-content/uploads/2020/12/CASDA-Language-Guide.pdf>

- Chenausky, K., Norton, A., Tager-Flusberg, H., and Schlaug, G. (2016). Auditory-Motor Mapping Training: Comparing the Effects of a Novel Speech Treatment to a Control Treatment for Minimally Verbal Children with Autism. *Plos one*, 11(11), e0164930. <https://doi.org/10.1371/journal.pone.0164930>
- Chiang, H-M. (2009). Differences between spontaneous and elicited expressive communication in children with autism. *Research in autism spectrum disorders*, 3(1), 214– 222. <https://doi.org/10.1016/j.rasd.2008.06.002>
- Crawford, M. J., Gold, C., Odell-Miller, H., Thana, L., Faber, S., Assmus, J., Bieleninik, L., Geretsegger, M., Grant, C., Maratos, A., Sandford, S., Claringbold, A., McConachie, H., Maskey, M., Mössler, K.A., Ramchandani, P., and Hassiotis, A. (2017). International multicentre randomised controlled trial of improvisational music therapy for children with autism spectrum disorder: TIME-A study. *Health technology assessment*, 21(59), 1-40. <https://doi.org/10.3310/hta21590>
- Dunn, L. M., and Dunn, D. M. (2007). *PPVT-4: Peabody picture vocabulary test- fourth edition*. Pearson Assessments.
- Fossum, K. L., Williams, L., Garon, N., Bryson, S. E., and Smith, I. M. (2018). Pivotal response treatment for preschoolers with autism spectrum disorder: Defining a predictor profile. *Autism research*, 11(1), 153–165. <https://doi.org/10.1002/aur.1859>
- Friedman, L., Sterling, A., DaWalt, L. S., and Mailick, M. R. (2019). Conversational language is a predictor of vocational independence and friendships in adults with ASD. *Journal of autism and developmental disorders*, 49(10), 4294-4305. <https://doi.org/10.1007/s10803-019-04147-1>

- Geretsegger, M., Elefant, C., Mössler, K. A., and Gold, C. (2014). Music therapy for people with autism spectrum disorder. *Cochrane database of systematic reviews*, (6).  
<https://doi.org/10.1002/14651858.CD004381.pub3>
- Girolametto, L., Verbey, M., and Tannock, R. (1994). Improving joint engagement in parent-child interaction: An intervention study. *Journal of early intervention*, 18(2), 155-167.  
<https://doi.org/10.1177/105381519401800204>
- Gordon, K., Pasco, G., McElduff, F., Wade, A., Howlin, P., and Charman, T. (2011). A communication-based intervention for nonverbal children with autism: What changes? Who benefits?. *Journal of consulting and clinical psychology*, 79(4), 447-457.  
<https://doi.org/10.1037/a0024379>
- Guerrero, N., Turry, A., Geller, D. and Raghavan, P. (2014). From historic to contemporary: Nordoff-Robbins music therapy in collaborative interdisciplinary rehabilitation. *Music therapy perspectives*, 32, 38–46. <https://doi.org/10.1093/mtp/miu014>
- Howlin, P., Goode, S., Hutton, J., and Rutter, M. (2004). Adult outcome for children with autism. *Journal of child psychology and psychiatry*, 45(2), 212-229.  
<https://doi.org/10.1111/j.1469-7610.2004.00215.x>
- Hume, K., Steinbrenner, J. R., Odom, S. L., Morin, K. L., Nowell, S. W., Tomaszewski, B., Szendrey, S., McIntyre, N. S., Yücesoy-Özkan, S., and Savage, M. N. (2021). Evidence-based practices for children, youth, and young adults with autism: Third generation review. *Journal of autism and developmental disorders*, 51(11), 4013–4032.  
<https://doi.org/10.1007/s10803-020-04844-2>

- Itzhak, E. B., and Zachor, D. A. (2011). Who benefits from early intervention in autism spectrum disorders?. *Research in autism spectrum disorders*, 5(1), 345-350. <https://doi.org/10.1016/j.rasd.2010.04.018>
- Janzen, T. B., and Thaut, M. H. (2018). Rethinking the role of music in the neurodevelopment of autism spectrum disorder. *Music and science*, 1, 1-18. <https://doi.org/10.1177/2059204318769639>
- Kasari, C., Gulsrud, A. C., Wong, C., Kwon, S., and Locke, J. (2010). Randomized controlled caregiver mediated joint engagement intervention for toddlers with autism. *Journal of autism and developmental disorders*, 40(9), 1045-1056. <https://doi.org/10.1007/s10803-010-0955-5>
- Kasari, C., Brady, N., Lord, C., and Tager-Flusberg, H. (2013). Assessing the minimally verbal school-aged child with autism spectrum disorder. *Autism research*, 6(6), 479-493. <https://doi.org/10.1002/aur.1334>
- Kasari, C., Kaiser, A., Goods, K., Nietfeld, J., Mathy, P., Landa, R., Murphy, S., and Almirall, D. (2014). Communication interventions for minimally verbal children with autism: a sequential multiple assignment randomized trial. *Journal of the American academy of child and adolescent psychiatry*, 53(6), 635–646. <https://doi.org/10.1016/j.jaac.2014.01.019>
- Kjelgaard, M. M., and Tager-Flusberg, H. (2001). An investigation of language impairment in autism: Implications for genetic subgroups. *Language and cognitive processes*, 16(2-3), 287-308. <https://doi.org/10.1080/01690960042000058>
- Koegel, L. K., Bryan, K., Su, P. L., Vaidya, M., and Camarata, S. (2019). Intervention for Non-verbal and Minimally-Verbal Individuals with Autism: A Systematic Review.

- International journal of pediatric research*, 5(2). <https://doi.org/10.23937/2469-5769/1510056>
- Koegel, L. K., Bryan, K. M., Su, P. L., Vaidya, M., and Camarata, S. (2020). Definitions of Nonverbal and Minimally Verbal in Research for Autism: A Systematic Review of the Literature. *Journal of autism and developmental disorders*. 50(8), 2957-2972. <https://doi.org/10.1007/s10803-020-04402-w>
- Koo, T. K., and Li, M. Y. (2016). A guideline of selecting and reporting intraclass correlation coefficients for reliability research. *Journal of chiropractic medicine*, 15(2), 155-163. <https://doi.org/10.1016/j.jcm.2016.02.012>
- Kover, S. T., and Atwood, A. K. (2013). Establishing equivalence: Methodological progress in group-matching design and analysis. *American journal on intellectual and developmental disabilities*, 118(1), 3-15. <https://doi.org/10.1352/1944-7558-118.1.3>
- Latif, N., Di Francesco, C., Custo-Blanch, M., Hyde K., Sharda M., and Nadig A. (2021). Joint engagement and movement: Active ingredients of a music-based intervention with school-age children with autism. *Neurorehabilitation*, 48(2), 167-185. [10.3233/NRE-208012](https://doi.org/10.3233/NRE-208012)
- Leonard, M. A., Milich, R., and Lorch, E. P. (2011). The role of pragmatic language use in mediating the relation between hyperactivity and inattention and social skills problems. *Journal of speech, language, and hearing research*. [https://doi.org/10.1044/1092-4388\(2010/10-0058\)](https://doi.org/10.1044/1092-4388(2010/10-0058))
- Lombardo, M. V., Pierce, K., Eyler, L. T., Carter Barnes, C., Ahrens-Barbeau, C., Solso, S., Campbell, K., and Courchesne, E. (2015). Different functional neural substrates for good and poor language outcome in autism. *Neuron*, 86(2), 567-577. <https://doi.org/10.1016/j.neuron.2015.03.023>



- Lord, C., Rutter, M., DiLavore, P.C., Risi, S., Gotham, K., Bishop, S. (2001). *Autism diagnostic observation schedule (ADOS)*. Los Angeles, CA: Western Psychological Services.
- Lord, C., Risi, S., and Pickles, A. (2004). *Trajectory of language development in autistic spectrum disorders*. In M. L. Rice and S. F. Warren (Eds.), *Developmental language disorders: From phenotypes to etiologies* (pp. 7–29). Lawrence Erlbaum Associates Publishers.
- Lord, C., Rutter, M., DiLavore, P. C., Risi, S., Gotham, K., and Bishop, S. (2012). *Autism diagnostic observation schedule—second edition (ADOS-2)*. Los Angeles: Western Psychological Services.
- Miller, J., Andriacchi, K., and Nockerts, A. (2016). Assessing language production using SALT software: A clinician’s guide to language sample analysis (PDF Edition). SALT Software
- Mössler, K., Gold, C., Aßmus, J., Schumacher, K., Calvet, C., Reimer, S., Iversen, G., and Schmid, W. (2019). The therapeutic relationship as predictor of change in music therapy with young children with Autism Spectrum Disorder. *Journal of autism and developmental disorders*, 49(7), 2795–2809. <https://doi.org/10.1007/s10803-017-3306-y>
- National Autism Center. (2015). *Findings and conclusions: national standards project, phase 2*. Randolph, MA
- Norbury, C. F. (2014). Practitioner review: Social (pragmatic) communication disorder conceptualization, evidence and clinical implications. *Journal of child psychology and psychiatry*, 55(3), 204-216. <https://doi.org/10.1111/jcpp.12154>
- Nordoff, P. and Robbins, C. (2007). *Creative Music Therapy: A guide to fostering clinical musicianship*. Barcelona Publishers: Gilsum, NH.

- Paul, R., Campbell, D., Gilbert, K., and Tsiouri, I. (2013). Comparing spoken language treatments for minimally verbal preschoolers with autism spectrum disorders. *Journal of autism and developmental disorders*, 43(2), 418–431. <https://doi.org/10.1007/s10803-012-1583-z>
- Pecukonis, M., Skwerer, D. P., Eggleston, B., Meyer, S., and Tager-Flusberg, H. (2019). Concurrent social communication predictors of expressive language in minimally verbal children and adolescents with autism spectrum disorder. *Journal of autism and developmental disorders*, 49(9), 3767-3785. <https://doi.org/10.1007/s10803-019-04089-8>
- Pickles, A., Anderson, D. K., and Lord, C. (2014). Heterogeneity and plasticity in the development of language: A 17-year follow-up of children referred early for possible autism. *Journal of child psychology and psychiatry*, 55(12), 1354-1362. <https://doi.org/10.1111/jcpp.12269>
- R Core Team (2013). R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. URL <http://www.R-project.org/>.
- Rose, V., Trembath, D., Keen, D., and Paynter, J. (2016). The proportion of minimally verbal children with autism spectrum disorder in a community-based early intervention programme. *Journal of intellectual disability research*, 60(5), 464-477. <https://doi.org/10.1111/jir.12284>
- Sandiford, G. A., Mainess, K. J., & Daher, N. S. (2013). A pilot study on the efficacy of melodic based communication therapy for eliciting speech in nonverbal children with autism. *Journal of autism and developmental disorders*, 43(6), 1298-1307. <https://doi.org/10.1007/s10803-012-1672-z>
- Särkämö, T., Altenmüller, E., Rodríguez-Fornells, A., and Peretz, I. (2016). Music, brain, and rehabilitation: emerging therapeutic applications and potential neural mechanisms. *Frontiers in human neuroscience*, 10, 103. <https://doi.org/10.3389/fnhum.2016.00103>

- Semel, E., Wiig, E., Secord, W., and Sample, N. (2004). Clinical evaluation of language fundamentals- fourth edition (CELF-4). San Antonio, Texas: *Psychological Corporation*.
- Sharda, M., Midha, R., Malik, S., Mukerji, S., and Singh, N. C. (2015). Fronto-temporal connectivity is preserved during sung but not spoken word listening, across the autism spectrum. *Autism research*, 8(2), 174-186. <https://doi.org/10.1002/aur.1437>
- Sharda, M., Tuerk, C., Chowdhury, R., Jamey, K., Foster, N., Custo-Blanch, M., Tan, M., Nadig, A., and Hyde, K. (2018). Music improves social communication and auditory-motor connectivity in children with autism. *Translational psychiatry*, 8(1), 231. <https://doi.org/10.1038/s41398-018-0287-3>
- Sharda, M., Silani, G., Specht, K., Tillmann, J., Nater, U., and Gold, C. (2019). Music therapy for children with autism: investigating social behaviour through music. *The Lancet child and adolescent health*, 3(11), 759-761. [https://doi.org/10.1016/S2352-4642\(19\)30265-2](https://doi.org/10.1016/S2352-4642(19)30265-2)
- Sihvonen, A. J., Särkämö, T., Leo, V., Tervaniemi, M., Altenmüller, E., and Soinila, S. (2017). music-based interventions in neurological rehabilitation. *The Lancet neurology*, 16(8), 648-660. [https://doi.org/10.1016/s1474-4422\(17\)30168-0](https://doi.org/10.1016/s1474-4422(17)30168-0)
- Srinivasan, S. M., & Bhat, A. N. (2013). A review of “music and movement” therapies for children with autism: embodied interventions for multisystem development. *Frontiers in integrative neuroscience*, 7, 22. <https://doi.org/10.3389/fnint.2013.00022>
- Tager-Flusberg, H., Rogers, S., Cooper, J., Landa, R., Lord, C., and Paul, R. et al. (2009). Defining Spoken Language Benchmarks and Selecting Measures of Expressive Language Development for Young Children With Autism Spectrum Disorders. *Journal of speech, language, and hearing research*, 52(3), 643-652. [https://doi.org/10.1044/1092-4388\(2009/08-0136\)](https://doi.org/10.1044/1092-4388(2009/08-0136))

- Tager-Flusberg, H., and Kasari, C. (2013). Minimally verbal school-aged children with autism spectrum disorder: The neglected end of the spectrum. *Autism research*, 6(6), 468-478. <https://doi.org/10.1002/aur.1329>
- Templin, M. C. (1957). *Certain language skills in children: Their development and interrelationships*. University of Minnesota Press. <https://doi.org/10.5749/j.ctttv2st>
- Thompson, G. A., McFerran, K. S., and Gold, C. (2014). Family-centred music therapy to promote social engagement in young children with severe autism spectrum disorder: A randomized controlled study. *Child: care, health and development*, 40(6), 840-852. <https://doi.org/10.1111/cch.12121>
- Thompson, G. A., and Elefant, C. (2019). “But I want to talk to you!” Perspectives on music therapy practice with highly verbal children on the autism spectrum. *Nordic journal of music therapy*, 28(4), 347-359. <https://doi.org/10.1080/08098131.2019.1605616>
- Timler, G. R. (2014). Use of the Children's Communication Checklist—2 for Classification of Language Impairment Risk in Young School-Age Children With Attention-Deficit/Hyperactivity Disorder. *American journal of speech-language pathology*. [https://doi.org/10.1044/1058-0360\(2013/12-0164\)](https://doi.org/10.1044/1058-0360(2013/12-0164))
- Trembath, D., and Vivanti, G. (2014). Problematic but predictive: Individual differences in children with autism spectrum disorders. *International journal of speech-language pathology*, 16(1), 57-60. <https://doi.org/10.3109/17549507.2013.859300>
- Trembath, D., Paynter, J., Sutherland, R., and Tager-Flusberg, H. (2019). Assessing Communication in Children with Autism Spectrum Disorder Who Are Minimally Verbal. *Current developmental disorders reports*, 6(3), 103-110. <https://doi.org/10.1007/s40474-019-00171-z>

- Venter, A., Lord, C., and Schopler, E. (1992). A follow-up study of high-functioning autistic children. *Journal of child psychology and psychiatry*, 33(3), 489-597. <https://doi.org/10.1111/j.1469-7610.1992.tb00887.x>
- Vivanti, G., Prior, M., Williams, K., and Dissanayake, C. (2014). Predictors of outcomes in autism early intervention: why don't we know more?. *Frontiers in pediatrics*, 2, 58. <https://doi.org/10.3389/fped.2014.00058>
- Wan, C. Y., Bazen, L., Baars, R., Libenson, A., Zipse, L., Zuk, J., Norton, A., and Schlaug, G. (2011). Auditory-motor mapping training as an intervention to facilitate speech output in non-verbal children with autism: a proof of concept study. *Plos one*, 6(9), e25505. <https://doi.org/10.1371/journal.pone.0025505>
- Wan, C. Y., and Schlaug, G. (2010). Neural pathways for language in autism: the potential for music-based treatments. *Future neurology*, 5(6), 797-805. <https://doi.org/10.2217/fnl.10.55>
- Wechsler, D. (1999). Wechsler Abbreviated Scale of Intelligence. *New York, NY: The Psychological Corporation, Harcourt Brace and Company.*
- Wechsler, D. (2003). Wechsler intelligence scale for children-fourth edition. *Bloomington, MN: Pearson.*
- Wechsler, D. (2014). Wechsler intelligence scale for children-fifth edition. *Bloomington, MN: Pearson.*
- Wong, C., Odom, S., Hume, K., Cox, A., Fettig, A., and Kucharczyk, S. et al. (2015). Evidence-Based Practices for Children, Youth, and Young Adults with Autism Spectrum Disorder: A Comprehensive Review. *Journal of autism and developmental disorders*, 45(7), 1951-1966. <https://doi.org/10.1007/s10803-014-2351-z>

- Yoder, P., and Stone, W. L. (2006). A randomized comparison of the effect of two prelinguistic communication interventions on the acquisition of spoken communication in preschoolers with ASD. *Journal of speech, language, and hearing research*. 49(4), 698-711.  
[https://doi.org/10.1044/1092-4388\(2006/051\)](https://doi.org/10.1044/1092-4388(2006/051))
- Yoder, P., Watson, L. R., and Lambert, W. (2015). Value-added predictors of expressive and receptive language growth in initially nonverbal preschoolers with autism spectrum disorders. *Journal of autism and developmental disorders*, 45(5), 1254-1270.  
<https://doi.org/10.1007/s10803-014-2286-4>

Figure 1

*Analysis 1 - Change in Coordinated Joint Engagement as a Function of Intervention Group and Verbal IQ (n = 42)*

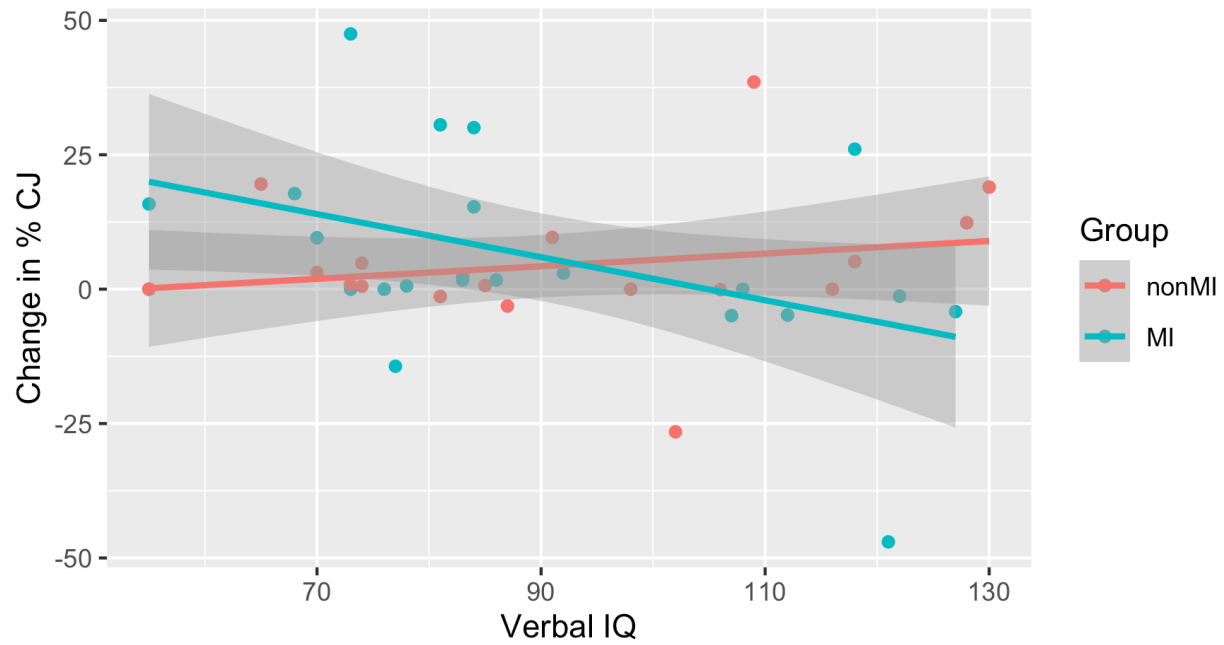
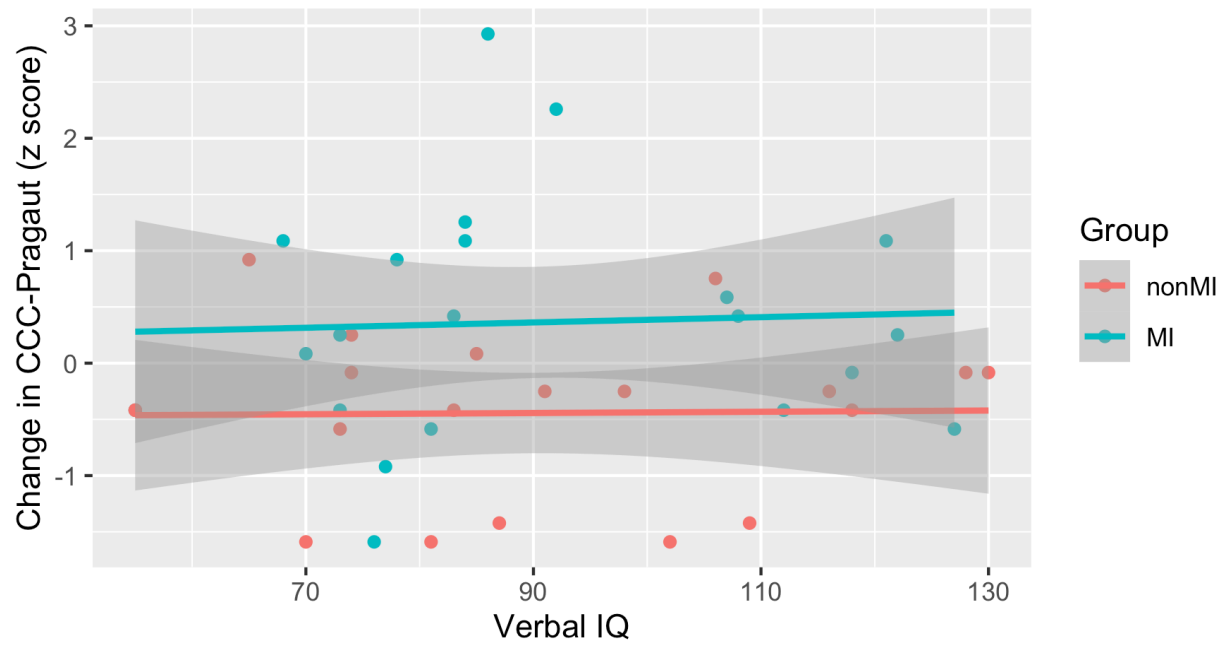


Figure 2

*Analysis 1 – Change in CCC-Pragmat as a Function of Intervention Group and Verbal IQ (n = 42)*

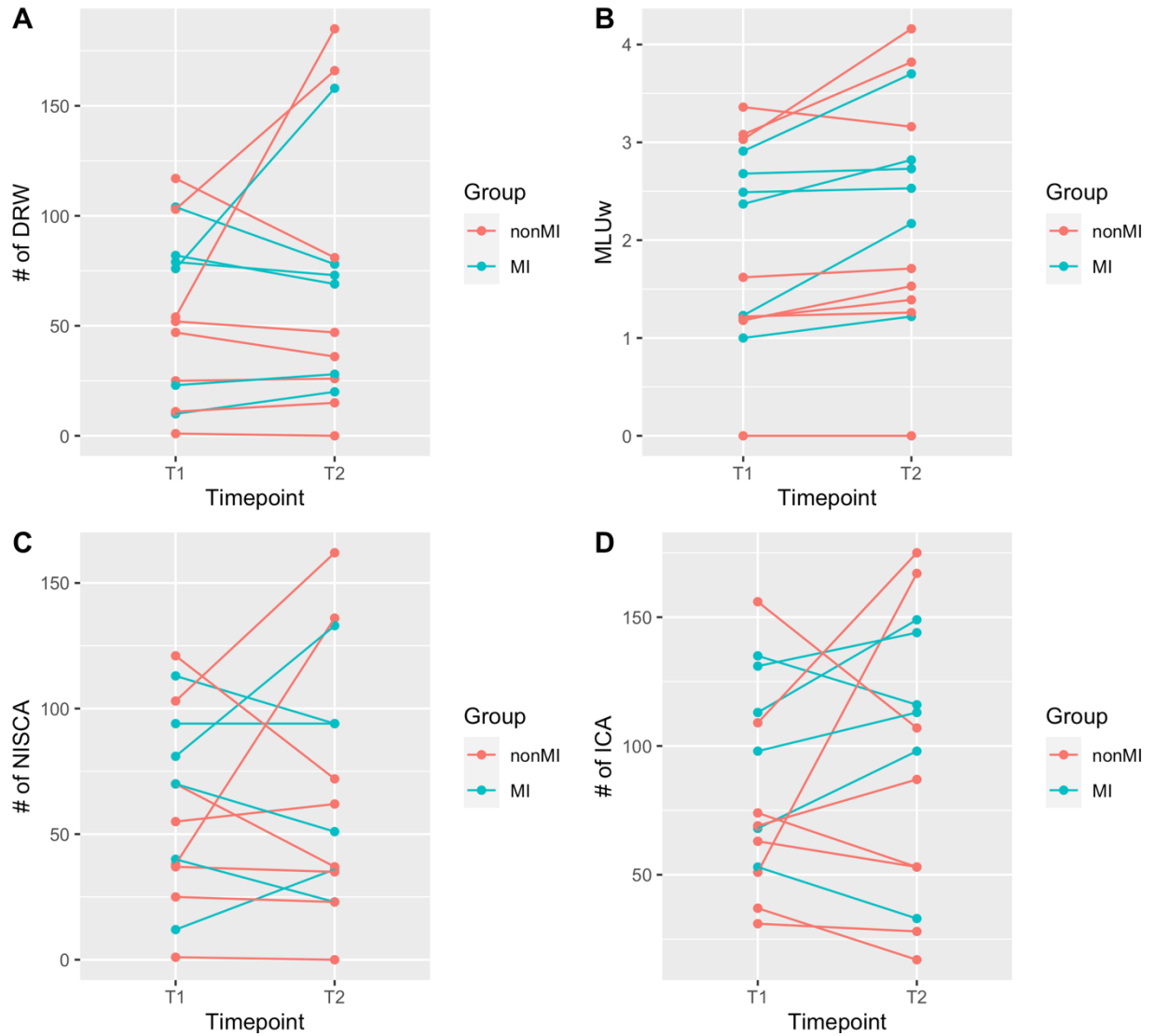




Running head: RESPONSE TO MUSIC-MEDIATED INTERVENTION IN AUTISTIC CHILDREN WITH LIMITED SPOKEN LANGUAGE

Figure 3

*Analysis 2 – NLS Variables as a Function of Timepoint in the Limited Spoken Language Ability Subsample (n = 14)*



# Running head: RESPONSE TO MUSIC-MEDIATED INTERVENTION IN AUTISTIC CHILDREN WITH LIMITED SPOKEN LANGUAGE

Table 1

*Analysis 1 - Participant Characteristics and Outcome Measures at Baseline for Main Sample (n = 42)*

	MI group <sup>a</sup>			nonMI group <sup>b</sup>					
<i>Participant Characteristics</i>	<i>n</i>	<i>Mean</i>	<i>SD</i>	<i>n</i>	<i>Mean</i>	<i>SD</i>	<i>p</i>	<i>Cohen's d/</i>	<i>Variance</i>
							<i>value</i>	<i>Chi-Square</i>	<i>Ratio</i>
Age (years)	21	10.6	1.9	21	10.1	2.0	0.50	0.27	1.11
Sex (male: female)	21	16:5	-	21	19:2	-	0.20	1.54 <sup>j</sup>	-
Autism Symptoms <sup>c</sup>	21	70.1	9.8	21	72.8	11.4	0.54	0.25	1.35
Performance IQ <sup>d,e</sup>	19	105.3	16.0	17	103.6	16.3	0.22	0.10	1.04
Verbal IQ <sup>d,e</sup>	20	92.0	19.67	19	91.84	21.7	0.20	<0.01	1.22
% Language Impairment <sup>f</sup>	19	58	-	22	53	-	0.66	0.11 <sup>j</sup>	-
Receptive Vocabulary Ability <sup>g</sup>	21	89.2	24.2	21	89.3	28.4	0.34	<0.01	1.38
<i>Outcome Measures at Baseline</i>									
% Time Spent per Activity in Coordinated Joint Engagement <sup>h</sup>	21	12.1	14.1	21	10.3	10.1	0.27	0.15	1.95
CCC-Pragmat Composite <sup>i</sup>	20	32.2	12.3	19	32.5	13.6	0.80	0.02	1.22

MI group: music-mediated intervention group, b) nonMI: non-music-mediated intervention control group, c) SRS-2: Social Responsiveness Scale, 2<sup>nd</sup> Edition T-score: higher scores indicate poorer skills, d) Wechsler Abbreviated Scale of Intelligence, 2<sup>nd</sup> edition, e) Wechsler Intelligence Scale for Children, 4<sup>th</sup> & 5<sup>th</sup> editions: mean of 100 with SD of 15, f) Percent of participants meeting criteria for language impairment based on scaled scores 1 SD or greater below (=7) of the mean (=10) on Clinical Evaluation of Language Fundamentals, 4<sup>th</sup> edition sentence repetition subtest, g) Peabody Picture Vocabulary Test, 4<sup>th</sup> edition standard score, h) Percent time spent per activity in a coordinated joint engagement state at timepoint 1, i) Children's Communication Checklist, 2<sup>nd</sup> edition pragmatic and autism unstandardized composite score at timepoint 1 which is the sum of 6 subscales scaled scores (individual scaled score range: 1-16): lower scores indicate poorer skills, j) Chi-square statistic used, \*p < 0.05, \*\*p < 0.01, \*\*\*p < 0.001, Cohen's *d* small effect = 0.2, medium effect = 0.5, large effect = 0.8

Table 2

*Analysis 2 - Correlation Between NLS Variables Used For The Limited Spoken Language Ability*

*Subsample (n = 14)*

NLS Measure <sup>a</sup>	ICA	NISCA	DRW	MLU <sub>w</sub>
ICA <sup>b</sup>	-			
NISCA <sup>c</sup>	0.9	-		
DRW <sup>d</sup>	0.9	0.9	-	
MLU <sub>w</sub> <sup>e</sup>	0.6	0.6	0.8	-

a) NLS = natural language sample, b) ICA = intentional communication act, c) non-spoken intentional communication act, d) DRW = total number of different root words, e) mean length of utterance in words, 0 = no correlation, <0.3 = small correlation, <0.5 = moderate correlation, <1.0 = strong correlation, 1 = perfect correlation

Table 3

*Analysis 1 - Regression Table for Fitted Coordinated Joint Engagement Model*

Coefficient	$\hat{\beta}$	SE ( $\hat{\beta}$ )	$t$	$p$
Intercept	4.28	3.45	1.24	0.222
Intervention Group	1.63	4.82	0.34	0.738
Verbal IQ	0.12	0.16	0.75	0.457
Group:Verbal IQ	-0.52	0.23	-2.28	0.029*

Multiple  $R^2 = 0.1497$ , Adjusted  $R^2 = 0.081$ ,  $RSE = 15.43(df = 37)$ ,  $n = 42$ ,  $F = 2.172$ ,  $p = 0.1077$ ,  
\* $p < 0.05$

Table 4

*Analysis 1 - Regression Table for Fitted CCC-Pragaut Model*

Coefficient	$\hat{\beta}$	SE ( $\hat{\beta}$ )	$t$	$p$
Intercept	-0.44	0.21	-2.11	0.042*
Intervention Group	0.81	0.29	2.74	0.009**
Verbal IQ	< 0.01	< 0.01	0.06	0.95
Group:Verbal IQ	< 0.01	0.01	0.13	0.90

Multiple  $R^2 = 0.1702$ , Adjusted  $R^2 = 0.1029$ ,  $RSE = 0.9398(df = 37)$ ,  $n = 42$ ,  $F = 2.53$ ,  $p = 0.07206$ , \* $p < 0.05$ , \*\* $p < 0.01$

Running head: RESPONSE TO MUSIC-MEDIATED INTERVENTION IN AUTISTIC CHILDREN WITH LIMITED SPOKEN LANGUAGE

Table 5

*Analysis 2 - Participant Characteristics and Outcome Measures at Baseline for Participants in the Limited Spoken Language Ability Subsample (n = 14)*

Baseline Variables	LSLA-MI group <sup>a</sup> (n=6)		LSLA-nonMI group <sup>b</sup> (n=8)				
<i>Participant Characteristics</i>	<i>Mean</i>	<i>SD</i>	<i>Mean</i>	<i>SD</i>	<i>p</i> value	<i>Cohen's d/</i> <i>Chi-square</i>	<i>Variance</i> <i>Ratio</i>
Age (years)	10.5	1.8	11.4	0.8	0.27	0.65	5.06
Sex (male: female)	4:2	-	7:1	-	0.35	0.88 <sup>j</sup>	-
Autism Symptoms <sup>c</sup>	77.3	7.8	74.1	11.7	0.55	0.32	2.25
Performance IQ <sup>d,e</sup>	100.3	16.8	92.1	18.6	0.42	0.46	1.23
Verbal IQ <sup>d,e</sup>	74.2	6.3	62.4	9.3	0.026*	1.49	2.18
<i>Outcome Measures at Baseline</i>							
ICA <sup>f</sup>	99.7	33.4	73.8	41.1	0.22	0.89	1.51
NSICA <sup>g</sup>	68.3	36.9	56.3	40.2	0.57	0.31	1.19
DRW <sup>h</sup>	62.3	37.1	51.3	41.2	0.61	0.28	1.23
MLUw <sup>i</sup>	2.11	0.80	1.84	1.19	0.61	0.27	2.21

a) LSLA-MI group: limited spoken language ability music-mediated intervention group, b) LSLA-nonMI: limited spoken language ability non-music-mediated intervention control group, c) SRS-2: Social Responsiveness Scale, 2<sup>nd</sup> Edition T-score: higher scores indicate poorer skills, d) Wechsler Abbreviated Scale of Intelligence, 2<sup>nd</sup> edition, e) Wechsler Intelligence Scale for Children, 4<sup>th</sup> & 5<sup>th</sup> editions: mean of 100 with SD of 15, f) ICA = intentional communication act, g) NSICA = Non-spoken intentional communication act, h) DRW = total number of different root words, i) MLUw = mean of length of utterance in words, j) Chi-square statistic used, \*p < 0.05, \*\*p < 0.01, \*\*\*p < 0.001, Cohen's *d* small effect = 0.2, medium effect = 0.5, large effect = 0.8

**Supplemental Information:**

Response to Music-Based Intervention in Autistic Children with Limited Spoken Language  
Ability

Journal of Autism and Developmental Disorders

### Supplemental Information - Section 1

Using terms and definitions proposed by Koegel et al., (2020), we sought to categorize the LSLA subsample into three subgroups based on increasing severity of language difficulty: (1) *limited speaking*: children who have significantly less spoken language than their typically developing peers, (2) *minimally speaking*: children who have a total spoken vocabulary of less than 50 words and do not combine words, and (3) *non speaking*: children who are over 18 months and do not produce any spoken language. Using these definitions, we used the natural language sample intervention video data at Timepoint 1 to assign each participant to one of these three subgroups.

### Supplemental Table 1

*Analysis 2 - Classification of Limited Spoken Language Ability Subsample (n = 14) using Koegel et al., (2020)*

Baseline Variables	LSLA-MI group <sup>a</sup> (n=6)		LSLA-nonMI group <sup>b</sup> (n=8)		<i>p</i> value	<i>Chi-square</i>
<i>Participant Characteristics</i>	<i>Mean</i>	<i>SD</i>	<i>Mean</i>	<i>SD</i>		
# Non Speaking <sup>c</sup>	0	-	0	-	-	-
# Minimally Speaking <sup>d</sup>	2	-	4	-	0.87	0.03
# Limited Speaking <sup>e</sup>	4	-	4	-	0.35	0.88

a) LSLA-MI group: limited spoken language ability music-mediated intervention group, b) LSLA-nonMI: limited spoken language ability non-music-mediated intervention control group, c) participants with no spoken words based intervention video at timepoint 1, d) participants with 50 spoken words or less and mean length of utterance in words (MLUw) of less than 2 units based on intervention video at timepoint 1, e) participants with DRW of two standard deviations below typical age peers using Leedholm and Miller, 1994 norms and a MLUw of 3 or less units based on cut-off from Pecukonis et al. (2019) using intervention video at timepoint 1, \* $p < 0.05$ , \*\* $p < 0.01$ , \*\*\* $p < 0.001$



## Supplemental Information - Section 2

**Fig. S1. Intervention design:** Both interventions, Music and Non-Music consisted of 45-minutes sessions. Each session had a defined structure with a ‘hello’ greeting, followed by selection of 4 activities per session using a visual schedule. A session log was used to ensure that each child participated in each of 9-11 activities an equal number of times. The sessions ended with a ‘goodbye’ theme and ‘clean up’. Both interventions targeted similar skills with and without the use of music.

Intervention Design

Session Structure	Music OR Non-Music	Skills targeted
Hello Greeting	<ul style="list-style-type: none"> <li>8-12 weeks of 45 minute, one-on-one sessions</li> <li>Combination of child- and therapist-led interaction</li> <li>9-11 activities counterbalanced over sessions</li> </ul>	<b>Communication</b>  <b>Social reciprocity and turn-taking</b>  <b>Sensorimotor integration</b>  <b>Emotion regulation</b>
4 choice pictogram		
Activity 1		
Activity 2		
Activity 3		
Activity 4		
Clean up and Goodbye		

Table S1. Intervention activities

Goal	Music activity	Non-music activity
Verbal Communication	Harmonica, Singing, Pete-the-Cat Story board with song, Recorder,	Pete-the-Cat Story board without song, Finger Puppets, Story book, Play Doh
Reciprocal Social communication	Piano, Xylophone, Pete-the-Cat Story board with song, Djembe	Pete-the-Cat Story board without song, Finger Puppets, Egg Shakers, Darts, Textured Bean Bags
Fine motor skills	Piano, Xylophone, Egg Shakers, Drums	Darts, Play Doh, Lego, Finger puppets,
Multisensory integration	Drums, Handheld Percussion, Xylophone, Melodica	Bubbles, Play Doh, Jigsaw, Lego, Finger Puppets
Emotional regulation	Drums, Singing,	Finger Puppets, Play Doh, verbal interaction during activities

### **Supplemental Information - Section 3**

#### *WASI -II - Verbal IQ Measure*

The WASI-II (Wechsler Abbreviated Scale of Intelligence – 2<sup>nd</sup> Edition) verbal IQ measure is composed of two subtests: vocabulary and similarities. In the vocabulary subtest, participants are asked to label a picture and for more advanced items, participants are given a spoken word and asked to define it. In the similarities subtest, participants are asked to identify two pictures that have similar qualities. More advanced items require the participant to describe the similarities between two spoken words. If scores from the Wechsler Abbreviated Intelligence Scale 4<sup>th</sup> or 5<sup>th</sup> editions (WISC-IV/V; Wechsler, 2003; Wechsler, 2014) from the previous two years were available, its verbal IQ score was used instead of the WASI-II.

#### **Supplemental Information - Section 4**

##### *Missing Data Imputation*

There was a total of 13 missing data points for the verbal IQ, coordinated joint engagement, and CCC-Pragaut variables combined. Eight data points (two verbal IQ at pre-intervention & six CCC-Pragaut with three at pre-intervention and three at post-intervention) were missing due to participants having very limited spoken language skills (i.e., non or minimally speaking), prohibiting administration of these spoken language measures. In these cases, the lowest possible score for that particular measure was assigned (i.e., verbal IQ = 55, CCC-Pragaut scaled score = 1 for each subscale for a total score of 6). Four additional data points (i.e., one CCC-Pragaut & three coordinated joint engagement at post-intervention) were missing due to attrition from the original RCT (Sharda et al., 2018). As was done in Sharda et al., (2018), the intention-to-treat principle was applied whereby these missing data points were assigned the participant's pre-intervention score indicating no change. Finally, in some cases participants provided IQ scores from a previous assessment conducted in the past two years. In one of these cases, a verbal IQ subtest result was not available, only a full-scale IQ score. For this single data point, no imputation was carried out.

### **Supplemental Information - Section 5**

#### *Analysis 2 - Screening Process for the Limited Spoken Language Ability Subsample*

An initial screening was conducted by three undergraduate research assistants in order to identify participants, who were suspected as having limited spoken language ability, for later time-intensive NLS transcription and coding. Screening criteria were having little or no spoken language or using an average of three spoken words or less per utterance with little variation in the words used. Research assistants watched the first 20 minutes of the first intervention session for each participant. Twenty-seven participants met the initial screening criteria.

A second round of screening was conducted by the first author, a speech-language pathologist with ten years experience assessing children on the autism spectrum, using the same criteria as applied by the undergraduate research assistants. Eight participants were found not to meet the outlined criteria: three children demonstrated their full spoken language ability later in the first session as they became comfortable with the therapist, and five children who presented with difficulties in spoken language but upon closer inspection spoke in sentences of on average four words and used a greater variety of words. This resulted in a sample of 19 participants for Analysis 2.

## References

- Koegel, L. K., Bryan, K. M., Su, P. L., Vaidya, M., and Camarata, S. (2020). Definitions of Nonverbal and Minimally Verbal in Research for Autism: A Systematic Review of the Literature. *Journal of Autism and Developmental Disorders*. 50(8), 2957-2972. <https://doi.org/10.1007/s10803-020-04402-w>
- Leadholm, B. and Miller, J. (1992). Language sample analysis: The Wisconsin Guide. Madison, WI: *Wisconsin Dept. of Public Instruction*.
- Pecukonis, M., Skwerer, D. P., Eggleston, B., Meyer, S., and Tager-Flusberg, H. (2019). Concurrent social communication predictors of expressive language in minimally verbal children and adolescents with autism spectrum disorder. *Journal of autism and developmental disorders*, 49(9), 3767-3785. <https://doi.org/10.1007/s10803-019-04089-8>
- Sharda, M., Tuerk, C., Chowdhury, R., Jamey, K., Foster, N., Custo-Blanch, M., Tan, M., Nadig, A., and Hyde, K. (2018). Music improves social communication and auditory-motor connectivity in children with autism. *Translational Psychiatry*, 8(1), 231. <https://doi.org/10.1038/s41398-018-0287-3>
- Wechsler, D. (2003). Wechsler intelligence scale for children-fourth edition. *Bloomington, MN: Pearson*.
- Wechsler, D. (2014). Wechsler intelligence scale for children-fifth edition. *Bloomington, MN: Pearson*.