Critical Review: Beyond Adults with Aphasia – The Effectiveness of Intonation-Based Therapy in Pediatric Populations

Taylor Riemersma
M.Cl.Sc SLP Candidate
University of Western Ontario: School of Communication Sciences and Disorders

Intonation-based therapy such as Melodic Intonation Therapy has been used to facilitate speech production in adults with aphasia for nearly thirty years. This therapy approach incorporates multiple mechanisms that might make it applicable to other populations. This critical review explores the effectiveness of intonation-based therapies to facilitate speech production in two pediatric populations: children with developmental apraxia of speech and children with autism associated with low verbal skills. A literature search yielded a total of eight articles, primarily single-subject designs as well as two case studies and two matched case-controls. Overall, there was suggestive evidence for the use of intonation-based therapy to establish early word productions in non-verbal or minimally-verbal children with autism. On the other hand, there was mixed evidence for the use of intonation-based therapy to increase intelligibility for children with apraxia. Recommendations for clinical practice and future research are discussed in this review.

Introduction

Melodic Intonation Therapy (MIT) was originally designed by Albert, Sparks & Helm in 1973 to facilitate propositional language in individuals with aphasia (Zumbansen, Peretz & Hebert, 2014). Broadly speaking, aphasia is a loss of speech and/or understanding of language as the result of left-hemisphere brain injury from a stroke or other brain insult. MIT was developed after clinicians and researchers observed intact singing abilities in this population that otherwise had little to no functional speech. In MIT, a patient melodically intones a target phrase while simultaneously tapping syllables with their left-hand to prime the motor cortex for speech.

Since singing primarily enlists the right hemisphere and not the left, this approach was thought to capitalize on non-injured language-capable brain regions in adults with aphasia (Zumbansen et al., 2014). However, recently it was proposed that MIT may have multiple underlying mechanisms working together (Merrett, Peretz & Wilson, 2014). That is, MIT may facilitate neural plasticity, engage the mirror neuron system, capitalize on the overlapping features of speech and music, and boost motivation and mood (Merrett, Peretz & Wilson, 2014). Since MIT may have multiple mechanisms involved, it is reasonable to assume that it could be applied to a broader range of populations beyond adults with aphasia. Two such populations are children with autism and children with developmental apraxia of speech (DAS).

Although there is no formal set of diagnoses criteria for DAS, this speech sound disorder is marked by three criteria: inconsistent errors on both vowels and consonants, disrupted transitions within and between words and abnormal prosody (American Speech-Language-Hearing Association, 2007). MIT could target these motor speech symptoms with its emphasis on rhythmic regularity, rate reduction and intensive practice (Zumbansen, Peretz & Hébert, 2014). In fact, MIT is most successful with Broca's aphasia patients whose distinguishing feature is apraxia of speech (Merrett, Peretz & Wilson, 2014; Zumbansen, Peretz & Hébert, 2014). It is important to consider the potential of MIT with this population since there is limited research on effective interventions for apraxia given the lack of formalized diagnostic criteria.

Another potential application is with children with autism. Although they are primarily characterized by deficits in social communication and social interaction (American Psychiatric Association, 2013), they may also have associated expressive and/or receptive language difficulties. As many as 30-50% of children with autism will not develop functional speech (Wan, Demaine, Zipse, Norton, & Schlaug, 2010). In addition, children with autism often experience oral motor difficulties (McCleery et al., 2013; Belmonte et al., 2013).

There are several reasons to believe that intonation-based therapy may be an effective therapy to improve speech production in children with autism. First, it might facilitate expressive language by strengthening neural networks between motor speech and language comprehension regions of the brain. This is an observed neurological deficit in children with autism (Wan & Schlaug, 2010), and it is this pathway (the arcuate

fasciculus) that has shown white matter changes after MIT in adults with aphasia (Schlaug, Marchina & Norton, 2009). Secondly, the multimodal approach of AMMT may also engage the mirror neuron system which is one proposed underlying deficit in autism and important for speech development (McCleery et al., 2013; Merrett, Peretz & Wilson, 2014). Further, since children with autism often have above-average pitch perception abilities, using intoned speech in therapy could target their strengths (Wan et al., 2010). For both pediatric populations, the potential of incorporating musical activity into speech therapy may boost engagement to encourage the repetitive practice necessary for success in treatment.

Objective

The purpose of this review is to determine if there is sufficient research to support the clinical use of intonation-based therapies (MIT and AMMT) with children with apraxia or autism to improve their speech production abilities and/or intelligibility.

Methods

Search Strategy

Online search engines and databases including Google Scholar, Scopus and CINAHL were searched using the following terms:
["Melodic Intonation Therapy" AND "child*"]
["Auditory Motor Mapping Training"].
The reference lists of relevant articles were also searched.

Selection Criteria

In order for a paper to be selected in this review, an intonation-based therapy (MIT or AMMT) had to be used with pediatric patients who did not have aphasia.

Data Collection

The search yielded a total of eight papers: four were single-subject design, two were case-control design and two reported individual cases.

Results

Results will be discussed in two sections: one on the application of intonation-based therapies with DAS and the other with autism.

Melodic Intonation Therapy & Developmental Apraxia of Speech

Single Subject Papers:

In a single-subject design, the participant or group complete both a treatment-free phase and an intervention phase to serve as their own control. This design is sensitive to how individuals respond to different treatments. However, without a true control, broad generalization cannot be made from this research. **Krauss and Galloway (1982)** explored if MIT would increase propositional language in children with apraxia as it does with adults with global aphasia. Participants included two children (ages not specified) with expressive language delay and apraxia whose profiles were similar to adults with aphasia, although participant diagnosis was sufficient to rule in oral but not necessarily verbal apraxia. Participants completed two months of traditional speech therapy as a control period followed by two months of MIT included as a warm-up before traditional speech therapy.

Outcome measures included appropriate formal and informal measures of expressive and receptive language including a composite involving intelligibility, but no separate measure of change in apraxia was included. Progress was evaluated at three points: before treatment as a baseline, after the initial control period and finally after the experimental MIT period. Appropriate statistical analyses demonstrated an increase in verbal naming ability, utterance length and intelligibility post-MIT.

This study's strengths include detailed language sample data and the use of a baseline. However, this study has several limitations including the diagnosis procedures and the lack of a separate apraxia change measure. Finally, no inter or intra-rater reliability for transcriptions was provided.

Overall, this study provides equivocal evidence regarding the use of MIT with children with DAS.

Martikainen (2011) explored the effectiveness of MIT and another motor-based intervention, Touch Cue Method (TCM), for children with apraxia. The one participant in this study was a Finnish girl (4 years; 7 months) with apraxia diagnosed according to appropriate criteria. She completed six weeks of MIT after a six-week baseline of no therapy. This was followed by another six-week treatment free period and finally six weeks of TCM. One element of the MIT was changed to substitute Finnish sign language for tapping of syllables. Data were collected at six points every six weeks and at a six-week follow up using trained and untrained stimuli in a picture naming task.

Well-described whole-word and segmental analyses were conducted on the participant's verbal naming productions. Strong inter-rater reliability of all phonetic transcriptions of targets was also reported. Appropriate statistical analyses indicated that on trained and untrained stimuli, the girl's percentage of vowels and words correct increased immediately after MIT and

consonant accuracy increased six weeks post-therapy. This improvement also continued during the TCM treatment period, especially for whole word accuracy. Overall, a decreased in glottalization was also noted.

This study's strengths include a well-described participant profile as well as detailed and appropriate analyses of outcome measures. However, this study is limited by a small sample, the use of a modified version of MIT and the lack of a control to rule out other factors contributing to the results.

Overall, this study provides a suggestive level of evidence for the use of MIT to improve articulation in children with DAS.

Lagasse (2012) compared the effects of standard speech language pathology versus MIT on speech production ability in children with apraxia. The participants in this study included two boys (five to six years old) with pure apraxia. Each child received alternating 40-minute sessions of each type of therapy for six weeks. While the MIT therapy is well-described, the control therapy was not sufficiently described. Pre- and post-test measures were conducted using two well-established formal articulation and phonology tests, however the measures were not sensitive to apraxia and not designed to be repeated within the three months. At the end of every therapy session (control or MIT) a speech production test specifically designed for this study was completed in the absence of the researchers.

Appropriate statistical analyses were used on the formal test scores and no significant improvements in articulation or phonology were found. Results on the speech production test were reported as trends. One participant had a slight improvement after MIT while the other's results were variable.

Therefore, this study provides equivocal support for MIT to improve speech production ability in children with apraxia.

Case Study Paper:
In clinical research, although case studies have poor external validity, they provide an in-depth exploration to encourage further research in that area.

Helfrich-Miller (1994) provided a clinical perspective on the use of MIT for children with DAS. After a description of the MIT program, case descriptions of three children with apraxia (two to eight years old) who completed MIT to improve articulation skills are outlined. A gold-standard measure of articulation, but not apraxia, was used to determine their initial phonetic inventories. This same measure was used to evaluate progress, but it was unclear how frequently. Each child received MIT for a different or unspecified length of time. One child did not receive MIT exclusively and it was unclear if the other two children received additional therapy as well.

No formal analyses were completed beyond a report of final phonetic inventories and informal observation. Results indicated that one child had a complete resolution of sound errors by age eleven and the other two children acquired all age-appropriate phonemes after several years of MIT.

Although the participant descriptions are detailed, this study has no consistently used appropriate outcome measures. The evidence from these case studies to support the use of MIT to improve articulation in children with apraxia is equivocal at best.

Melodic Intonation Therapy/Auditory Motor Mapping Training & Autism

Case Study:
Miller and Toca (1979) explored the first potential application of MIT for a child with autism. In this case study, a thorough case history was provided of one three-year old boy who received an adapted version of MIT that included signing while singing. The duration of his therapy was not reported.

No formal measures were used to qualify the stated purpose of the therapy. Examples of target words produced (e.g. "cookie") were given throughout to demonstrate his progress. Generalization of target words was also informally reported. Overall, without formal outcome data, the authors appeared to have made overextended claims regarding his progress and how MIT benefited him. The evidence provided in this case is equivocal at best.

Single Subject Papers:
Auditory Motor Mapping Training (AMMT) is a modified version of MIT that involves intoning target word or phrases while tapping the rhythm on tuned drums.

Wan and colleagues (2011) conducted this study to demonstrate the initial efficacy of AMMT in facilitating speech output in non-verbal children with autism. The participants included six non-verbal children (five male) between the ages of five and nine chosen according to appropriate selection and diagnostic criteria. They completed 40 sessions of AMMT after an initial no-treatment baseline and were followed-up four and eight weeks after therapy. The intervention is laid out in detail, down to the precise tuning of each drum.

Treatment fidelity was also randomly assessed throughout the study. While no statistic is reported, qualitative report lists the treatment adherence as high.

A probe was administered at baseline, every five sessions during treatment and at two follow-up points up to eight weeks later using trained and untrained stimuli. The probes were an appropriate level of syllabic difficulty for non-verbal participants. Transcriptions were completed by coders blinded to the probe session and high inter-rater and intra-rater reliability were reported. The rules for transcriptions are clearly laid out supporting replicability and sensitive to the verbal ability of the participants.

For each participant, descriptive statistics reporting the difference between best baseline and session forty confidence intervals were given. An appropriate statistical analysis indicated that each child's speech approximations of CV syllables improved after forty AMMT sessions and this improvement continued up to eight weeks after the last session.

Overall, this study provides suggestive support for the use of AMMT to improve speech output in non-verbal children with autism.

Matched Case-Control Design Papers:
In understanding efficacy of a treatment versus control in clinical populations, a matched control is often necessary. However, the lack of a true control limits external validity

Chenausky and colleagues (2016) explored the effect of AMMT compared to a control, Speech Repetition Therapy (SRT) for minimally-verbal children with autism. The participants included 21 minimally-verbal children with autism chosen with appropriate diagnostic and selection criteria. According to quasi-randomized assignment, thirteen received AMMT while eight children received the control. Seven from each group were matched based on age, mental age and an established measure of apraxia. The treatment structure was well-described and high treatment fidelity was reported.

Trained and untrained word-level stimuli used as outcome measures were reasonable targets for minimally verbal children. Although only moderate inter-rater reliability was reported, a reasonable rationale for these lower scores was provided. An appropriate statistical analysis indicated that the percentage of syllables, vowels and consonants correct on trained and untrained stimuli improved for the AMMT group after 25 sessions. There was also a significant difference between AMMT and control for percentage of syllables and consonants correct. Finally, more children responded in AMMT than in control therapy. Play and social skills were also informally observed to improve.

This study has several strengths with its matched control and detailed descriptions. However, the study's strict inclusion criteria may limit how well the results generalize to all children with autism. Overall, however this study provides a compelling level of evidence for the use of AMMT in minimally-verbal children with autism.

Chenausky and colleagues (2017) explored if AMMT would have similar effects in more-verbal children with autism as with minimally-verbal children. Two minimally-verbal children and two more-verbal children, all diagnosed with autism, participated in this study, for a total sample size of four participants, limiting external validity. Participants in each dyad were well-matched and selected according to appropriate diagnostic and inclusion criteria. In each dyad (more-verbal and minimally-verbal), one child received AMMT and the other a non-intonation based control, speech repetition therapy (SRT). Either bisyllabic or trisyllabic stimuli were used for the minimally-verbal versus more-verbal children to match their respective verbal abilities. Outcome measures are clearly described and conducted at baseline and post-25 sessions of AMMT. Inter-rater reliability for transcriptions was low and the rationalization provided may not have been appropriate for the participants' higher language levels.

Appropriate statistical analyses were conducted with baseline scores included as a covariate due to participant differences. AMMT resulted in a greater increase in syllables, consonants and vowels correct in both trained and untrained stimuli than the control therapy and these results were associated with large effect sizes. Logical rationale is provided for the lack of improvement seen on syllables approximated in more-verbal children with autism, attributing it to a measure that is insensitive to higher language levels.

Two major weaknesses with this study are its poor validity due to a small sample size and low levels of interrater reliability. Overall, this study provides a suggestive level of evidence that AMMT can successfully be applied to more-verbal children with autism to improve their speech production.

Discussion

This critical review explored the use of intonation-based therapy to improve speech production abilities

and intelligibility in children with disorders that may be associated with speech and/or language problems. Overall, across both disorder types stronger evidence emerged for the use of Auditory Motor Mapping Training (AMMT), a variation of Melodic Intonation Therapy (MIT), for non-verbal or minimally-verbal children with autism. On the other hand, results for the use of MIT with children with developmental apraxia of speech were inconsistent.

With regards to children with apraxia of speech, the evidence was largely equivocal. One more recent study was suggestive, but limited by an alternating design that could not separate treatment effects. However, given that significant improvements were seen in this study when MIT was used with another therapy, Touch Cue Method, this could indicate that MIT may be effective at improving speech production when used in conjunction with other established methods. Claims beyond improving word-level intelligibility cannot be made since any measurements of change in apraxia were at this level. Therefore, MIT may be more effective as an initial warm-up or stepping stone to other goals targeting meaningful change at conversational level.

While there was no evidence to support the use of MIT with children with autism, a stronger evidence based emerged to support the use of AMMT with children with autism and associated language delay. The overall highly suggestive research available demonstrated that AMMT could successfully improve speech approximations of target words in children with autism at varying language levels (i.e., minimally to more verbal). Not only does this research demonstrate efficacy of AMMT as a treatment, but that it could be applicable to a variety of autism profiles.

Several studies cited that without controls it could not be determined if intonation or repetitive practice was the contributing factor to positive results. However, one study on AMMT demonstrated that intonation is likely the critical component since the AMMT group speech production scores significantly surpassed any speech improvements in the control group. However, none of the studies exploring MIT for children with apraxia included controls. Future research in this area should do so to confirm that intonation is the critical component of change.

Another theme arising from the research is the question as to what the ideal and minimal length of effective treatment would be, especially for children with apraxia. Every study had a varied length of treatment from four sessions to several years. This wide range may have contributed to the mixed evidence seen for the use of MIT with children with DAS. For efficient clinical practice, future research should explore the minimal length of treatment necessary to produce significant changes.

Future Research Considerations:
For both pediatric populations, there is a limited research base on the efficacy of intonation-based therapies. Future research should include larger samples and well-designed controls to improve external validity. To broaden the scope of the treatment, stimuli should include targets at a phrase or sentence level or even alternative goals that included interaction skills. Future research should also include a variety of ages and disability levels. Finally, given that Melodic Intonation Therapy was originally designed to target propositional language rather than speech production, the use of Auditory Motor Mapping Training, a therapy specifically targeting intelligibility in speech production, might be more effective for children with apraxia and warrants exploration in future research.

Clinical Implications

Overall, due to poor study designs there is equivocal support for the use of intonation-based therapy for children with apraxia. Melodic Intonation Therapy (MIT) should be used for this population only with careful evaluation of ongoing progress and in conjunction with another established method. Based on an overall suggestive level of evidence, clinicians can feel more confident in the exclusive use of Auditory-Motor Mapping Training (AMMT) to establish early word productions in children with autism with low verbal skills. Overall, intonation-based therapy is a promising therapy for pediatric populations and should continue to be explored in clinical research and practice.

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