REVISTA CEFAC
SPEECH, LANGUAGE, HEARING SCIENCES AND EDUCATION JOURNAL
Rev. CEFAC. 2017 Mar-Abr; 19(2):289-298
doi: 10.1590/1982-021620171928516

Case reports

Verbal behavior teaching by multiple exemplars in a child with auditory neuropathy spectrum disorder: a case study

Ensino de comportamento verbal por múltiplos exemplares em uma criança com desordem do espectro da neuropatia auditiva: estudo de caso

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Data was obtained during the first author's Master's research under supervision of the fifth author. Since the participant did not fulfill the criteria for inclusion in the sample, the case was analyzed and described individually.

Conflict of interest: non-existent

Received on: July 05, 2016
Accepted on: January 18, 2017

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ABSTRACT

Auditory Neuropathy Spectrum Disorder (ANSD) is characterized by a difficulty in establishing verbal behavior in the speaker and listener modalities due to an asynchrony in the nervous conduction of sound stimuli, leading to hearing loss. Cochlear implants are an alternative that stabilizes sound detection; however, discriminating and recognizing what is heard and establishing relations to speech requires learning. A procedure with recognized efficacy in establishing verbal repertoires in children with little or no language is Multiple Exemplar Instruction (MEI). The objective of this study was to verify replicability of results obtained from MEI with other populations for teaching and integrating listener and speaker behaviors on a six year-old child with ANSD and bilateral cochlear implants. The study was conducted with three sets of stimuli and consisted in a rotated presentation of listener responses, based on figure selection from dictated words, and of speaker responses, after a dictated word (echoic) and figure naming (tact). Repeated probes evaluated performance on the set of stimuli. Even though there is some variability in responding, selection responses were established first and speaker responses occurred gradually throughout successive sessions. The conditions under which a speaker's repertoire may be refined, and generalization of these results to other participants with ANSD should be further investigated.

Keywords: Language; Hearing Impaired Persons; Cochlear Implant; Teaching

RESUMO

A Desordem do Espectro da Neuropatia Auditiva (DENA) é caracterizada pela dificuldade no estabelecimento do comportamento verbal nas modalidades de ouvinte e de falante, por uma dessincronia na condução nervosa da estimulação sonora recebida, ocasionando perdas auditivas. O implante coclear tem sido uma alternativa que estabiliza a detecção sonora, porém, discriminar e reconhecer o que se ouve e estabelecer relações com a fala irá requerer aprendizagem. Um procedimento de reconhecida eficácia no estabelecimento de repertórios verbais em crianças com pouca ou nenhuma linguagem é o *Multiple Exemplar Instruction (MEI)*. O objetivo deste estudo foi verificar a replicabilidade dos resultados obtidos com o MEI com outras populações no ensino e integração entre os repertórios de ouvir e de falar em uma criança de seis anos com DENA e implante coclear bilateral. O ensino foi conduzido com três conjuntos de estímulos, e consistiu na apresentação rotativa de respostas de ouvinte, baseadas na seleção de figuras mediante palavra ditada e, de falante, após uma palavra ditada (ecoico) e mediante a nomeação de uma figura (tato). Repetidas sondas avaliaram o desempenho nos conjuntos de estímulos. Ainda que com alguma variabilidade no responder, as respostas de seleção foram estabelecidas primeiro e as respostas de falante, gradativamente, ao longo de sucessivas sessões. As condições sob as quais o repertório de falante pode ser refinado e a generalização dos resultados para outros participantes com DENA devem ser investigadas.

Descritores: Linguagem; Pessoas com Deficiência Auditiva; Implante Coclear; Ensino

INTRODUCTION

Auditory Neuropathy Spectrum Disorder (ANSD) is clinically characterized as a difficulty in word comprehension for cases of little to moderate hearing loss, especially in noisy environments[1]. It was initially named Auditory Neuropathy (AN) or Auditory Dyssynchrony (AD)[2] due to its association to direct impairment of the cochlear nerve. However, since not all ANSD patients have a lesion in this nerve, the definition is now broader and includes other lesion loci that are responsible for electrical transmission and sinapses[3]. Considering the audiological characteristics, thresholds for sound detection may present varied configurations, from normal hearing to profound hearing loss, unilateral or bilateral, symmetrical or not. Additionally, occurrence of cases of progression and fluctuations in hearing loss is possible; in some of these cases, fluctuations may occur from one day to the next[4].

Early diagnosis is especially important, since this condition interferes with the development of language. Parents or healthcare professionals are usually the ones to raise suspicions. Diagnosis is conducted through audiometric and electrophysiological tests, and is confirmed by the presence of otoacoustic emissions (OAE) associated to altered or absent brainstem auditory evoked potential (BAEP). Tests are quick, non-invasive, easy to conduct, are approximately 100% accurate[1,3].

The cochlear implant (CI) is among the strategies indicated for auditory habilitation and rehabilitation especially for children in a critical period for developing linguistic skills as listeners or speakers. However, CIs are still a challenge for individuals with ANSD, since impairment of auditory perception, and speech comprehension and production, originate from alterations in neural functions[3,5]. The CI is an electronic device designed to promote auditory detection and, consequently, improve communication of people. It is used when a patient cannot benefit from prosthesis that prioritize amplifying sound, such as Personal Sound Amplification Products (PSAP). The CI is composed of an internal part, which is chirurgically implanted with electrodes inserted in the cochlea. The external part, a microphone installed near the ear, captures sound, which is transmitted through a wire to the speech processor. The processor sends the coded information to a transmission antennae placed near the stimulating receptor. The auditory cycle completes when the electrical stimulus and coded signals are transmitted through radio frequency to the receptor-transmitter. This apparatus stimulates the electrodes that are implanted in the cochlea[5,6].

The CI has been successful in developing cortical hearing and shown behavioral results from children with ANSD[3,7,8]. In general, children with ANSD that received the implant when they were less than two years old were most likely to present similar auditory results than their counterparts without ANSD and CI[7]. Moreover, there were no significant differences between children with ANSD and CI, assessed with different instruments, and children with sensorineural hearing loss, since evolution of skills was quite similar[3]. Considering phonoaudiologic systematic interventions, this seems like a good alternative to develop pragmatic communication skills in children with ANSD without CI[8]. Thus, it is possible to see that children with ANSD respond well to teaching programs.

However, there is also great variability in the literature regarding persons with ANSD and CI. This variability in results relates to extrinsic factors – such as motivation to use the CI, family support, rehabilitation method – and intrinsic factors – such as degree of impairment or changes in the auditory pathways[5]. Defining and standardizing clinical practices for a clinical group with ANSD is not yet possible considering its heterogeneity. Similarly, relevant teaching conditions to learn specific language repertoires must be investigated further.

Considering that persons with ANSD have a significant delay in the development of linguistic repertoires, as listeners as well as speakers, the present study has followed the performance of a child exposed to a procedure to teach these skills.

Linguistic repertoire categories have been described for a better understanding of speakers' behaviors due to its diversity based on events that precede responses and their consequences. Thus, these categories are identified independently from their topography, be it a vocal, motor, or visual (image swapping system) response. The focus is on verifying the controlling variables that precede each operant and its function in the environment[9]. The present work will target the categories "echoic" and "tact", and the listeners' behaviors[10]. Echoic behavior is usually present in initial stages of development and is characterized as vocal or motor responses that are controlled by an auditory verbal stimulus; there is a structural identity between the antecedent stimulus and the response[10]. Tact, on the other hand, is a vocal response that is controlled by the presence of a non-verbal stimulus, be it public or private (objects, events, feelings). A listener contacts what controls the verbal behavior of the speaker through tact[10]. A listener's behavior is hereby defined as responding differentially to different auditory stimuli.

Children with typical development usually acquire listener and speaker behaviors incidentally, so the independence relations between listening and speaking behaviors are practically imperceptible. Children with delayed language development show clearly that learning one type of linguistic repertoire can be maintained independently. In such cases, it is necessary to program direct teaching conditions to relate the lacking repertoire to those that have been previously established[11]. For example, picking up a glass of water when mother asks (listener behavior) is not a necessary condition for the child to later ask for water when he or she is thirsty (speaker behavior) or say "water" when looking at a glass of water. Research has shown that interdependence between different types of verbal behaviors require specific contingencies to be programmed[12-15].

Among the procedures used to establish interdependent relations between listening and speaking, Multiple Exemplar Instruction (MEI) has shown promising results with participants with minimal verbal repertoires, especially those with Autistic Spectrum Disorder[16-18]. MEI consists in rotating tasks that involve listener and speaker skills by considering different stimulus control on listening (dictated word, selection), speaking (dictated words for echoic behavior and naming figures for tact). MEI can produce shared stimulus control, between stimuli that were initially independent, through successive tasks[19]. Thus, if speaking is precise only for echoic tasks (repetition), it may also occur for tasks involving tact (figure naming) when teaching promotes rotation between these tasks. Even though efficacy of MEI is demonstrated in various studies conducted with autistic children, it has not been much studied with other populations with language delay. Thus, the present work had the objective of replicating positive results obtained with MEI with other populations for teaching and integrating listener and speaker repertoires to a child with ANSD and CI.

CASE PRESENTATION

Ethical procedures

Throughout the procedure all ethical considerations were made regarding the participants' rights according to the Ethics Committee for Research (CEP Protocol nº 440532). Authorization was obtained from the participant and her legal guardians, who received a Consent Form. The guardians were aware of the objectives of the study, their voluntary agreement to participate, the confidentiality of their identity, and the absence of onus to their participation; they agreed to disclosure of results only in academic and scientific contexts.

Participant

A single child, of female gender, aged six years old and nine months at the time of the intervention participated in this study. She had bilateral profound sensorineural hearing loss and Auditory Neuropathy Spectrum Disorder. The child had a bilateral cochlear implant; the first was implanted on the left ear five years before intervention and the second was implanted on the right side five months before the study. Auditory deprivation occurred for a period of three years and eight months.

According to the PPVT-IV *Peabody Picture Vocabulary Test – Revised*[20], which evaluated receptive vocabulary, the participant presented an age of two years and ten months. In the language category, measured by the Meaningful Auditory Integration Scale (MAIS)[21] she scored 5, on a scale from 0 to 6. In the hearing category, measured by the Meaningful Use of Speech Scales (MUSS)[22], she scored 3, on a scale from 0 to 5.

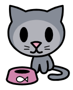
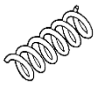
The participant was enrolled in the first year of Elementary School and, outside school hours, she went to a public health center with a complementary education focus that is directed to people with hearing loss.

Materials, Stimuli and Experimental Conditions

In this study we used an iBook G4 microcomputer with the MTS®[23] software for presentation of teaching tests and tasks. To record the participant's performance in speech production tasks, we used a Sony Cyber-shot DSC-W530 camera. Teaching and test tasks were conducted with discrete trials, in which two types of responses were required: speaker and listener. Listener responses consisted in selecting figures after an auditory stimulus was presented and speaker responses were echoic or tact.

Three sets of visual and sound stimuli were used. Table 1 shows the sets of stimuli used in the teaching and testing routines in the sound and visual modalities.

Table 1. Stimuli used in teaching and testing phases

Set 1		Set 2		Set 3	
Word	Picture	Word	Picture	Word	Picture
"PATO"		"MALA"		"FADA"	
"VACA"		"DADO"		"GATO"	
"MOLA"		"LATA"		"CASA"	

Sessions were conducted in a room in the institution where the child received treatment and only the participant and the experimenter were at the room during the sessions. Three types of trials were presented: echoic, selection and tact, as described below.

Echoic: The trials started with a blue square at the center of the computer screen; simultaneously, one of the auditory stimuli was emitted through the speakers, soliciting for the participant to repeat the dictated word. Once the child responded, the experimenter clicked on the blue square, which ended the trial and started the next one.

Figure selection: The trials started with the auditory stimulus through the speaker. One click to the blue square at the center of the screen produced two visual stimuli, which were presented randomly in two of the four corners of the screen. The participant had to mouse-click on the image that corresponded to the auditory stimulus and the trial ended.

Tact: Trials started with a figure on the center of the screen and the child was requested to name it. After emitting the oral response, a mouse-click on the figure ended the trial.

Training and testing trials were presented. Training trials ended with differential consequences for correct responses (colorful stars and a jingle) and incorrect responses (black screen). Test trials were not followed by consequences (a gray inter-trial interval screen was presented).

Procedure

Teaching was structured by MEI (*Multiple Exemplar Instruction*) and intercalated with tests of the same selection, echoic and tact relations, before and after each teaching set. Table 2 presents a summary of these stages.

Table 2. Teaching and testing structure used in the study

Test	Teaching	Test	Teaching	Test	Teaching	Test
Set 1	Set 1 (MEI)	Set 1		Set 1		Set 1
Set 2		Set 2	Set 2 (MEI)	Set 2		Set 2
- - -		Set 3		Set 3	Set 3 (MEI)	Set 3

MEI = Multiple Exemplar Instruction

Tests

Tests were carried out to evaluate the echoic, picture selection, and tact operants for the three sets of stimuli. Tasks were presented in blocks of nine trials: three echoic, three tact and three selection. A trial block was presented for each stimulus set. Responses emitted by the participant in these stages had no consequence, regardless of being correct or incorrect.

Teaching

Echoic, word selection and tact relations were taught for each set of words. The procedure consisted of teaching listener (selection of pictures) and speaker (echoic and tact) relations structured by MEI. The relations of a set of stimuli were presented in blocks of 27 trials, which consisted of nine selection trials, nine echoic trials and nine tact trials. Trials were presented in groups of three, consisting of one echoic, one selection and one tact trial of the same stimulus. For example, in the case of "*pato*" (duck), the participant asked to "Say /*pato*/", followed by "Point to /*pato*/", and finally "Which Picture is this? (Picture of a duck)". This procedure was repeated three times for each stimulus in a set, in random order. Correct responses were reinforced by a sticker, or a small piece of milk chocolate, as well as praises from the experimenter and the consequences of the teaching program. Incorrect responses were corrected, followed by a new opportunity to respond after the correction, according to the sample offered by the experimenter.

Data analysis procedure

In order to analyze the participant's responses for the echoic and tact operants, we conducted a phonemic evaluation (point-to-point correspondence) of the programmed word, and from the total of correct phonemes obtained the percentage of correct responses. For example, in order for the word PATO from set 1 to have a 100% correct score, the child needed to emit the four phonemes /p/-/a/-/t/-/o/. In case a different response was emitted, such as _/a/-/t/-/o/ (omission) or /b/-/a/-/t/-/o/ (distortion), the correct corresponding phonemes were counted, in this case three phonemes, totaling a 75% correct[24].

RESULTS

Figure 1 presents the participant's performance after being exposed to teaching by multiple exemplars until reaching the learning criterion.

Generally, for the three teaching sets, we observed that the listener repertoire, selecting pictures, was either well established (sets 1 and 3) or was the first to be established (set 2) even if teaching tasks for speaker behavior (echoic and tact) were presented. Considering speaker behaviors, the first to be acquired when learning the three sets was the echoic response. This was followed by an increase in the number of emissions of tact responses for the three sets, in relation to the base line.

During the teaching by multiple exemplars phase for set 1, the participant emitted the correct response for all nine trials in the listening block based on selection on the first exposure to the block. Regarding tact, although it was emitted with some correspondence (five correct responses in the first block), it did not vary much throughout the eight teaching blocks. However, echoic responses occurred with three correct responses in the first block and with eight correct responses in the last teaching block, which was close to the selection levels.

In the teaching phase for set 2, the number of correct responses obtained in the selection, echoic, and tact operants was below 4. Thirteen teaching blocks were required for the selection operant to reach nine correct responses. The correct responses obtained in the echoic and tact operants gradually increased until reaching six for echoic and four for tact.

For set 3, only four sessions were required for the participant to obtain nine correct responses in selection and eight for both echoic and tact.

The effect of teaching and successive probes is demonstrated in Figure 2, below. The dotted line delimits the moment in which each set was taught: to the left of the line are the performances for the target operants before teaching of the respective set; to the right are the results after teaching and monitoring of each.

It is possible to observe in Figure 2 that, after teaching set 1, the results for the selection operant remained at around 100%. For the echoic and tact operants the percentage of correct responses increased in relation to the pre-test by 41.6% for echoic and 50% for tact. Next, set 2 was tested again and, without training, echoic performance increased by 25% and tact by 16.7%.

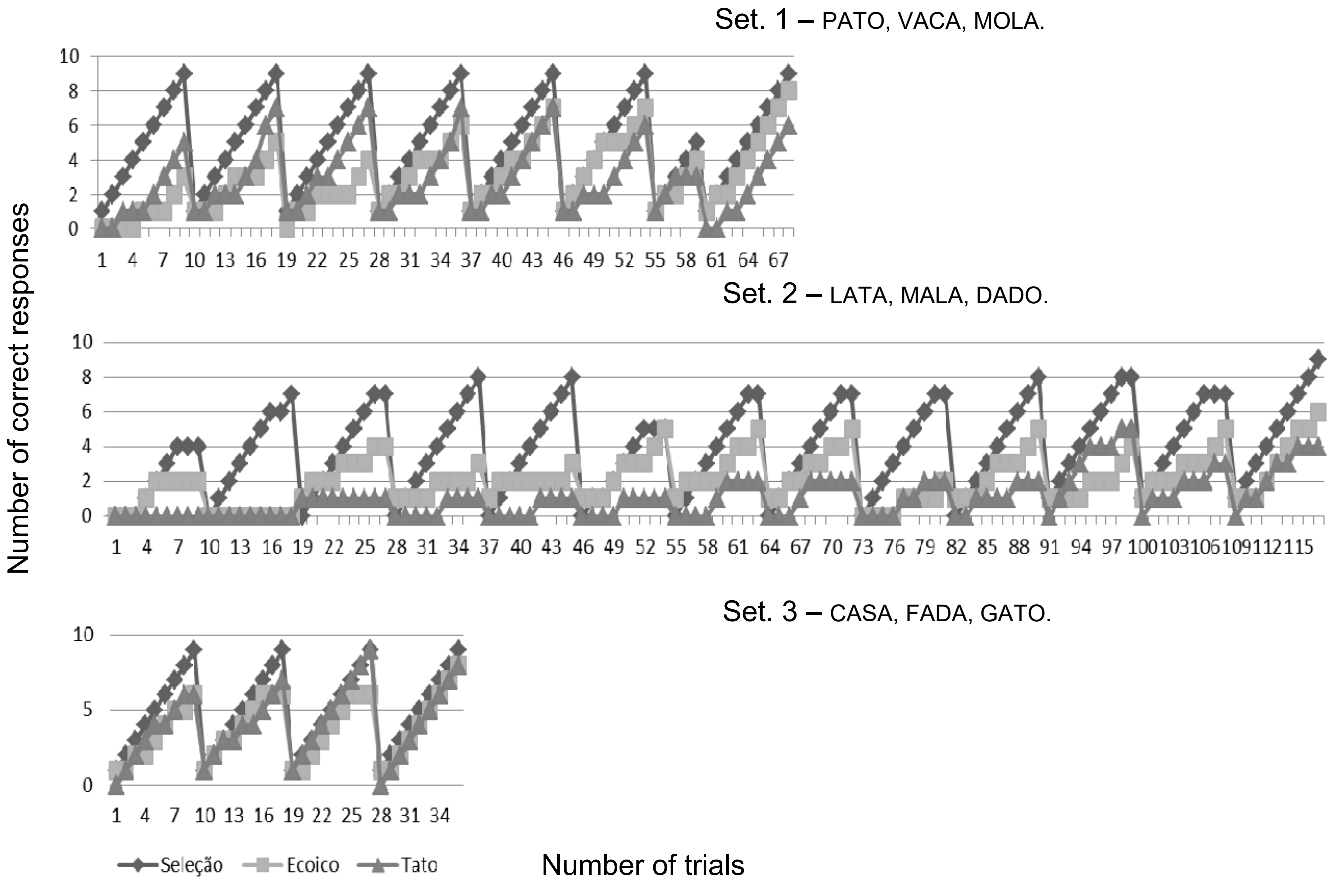


Figure 1. Performance of participant exposed to teaching with multiple exemplars until reaching the learning criterion

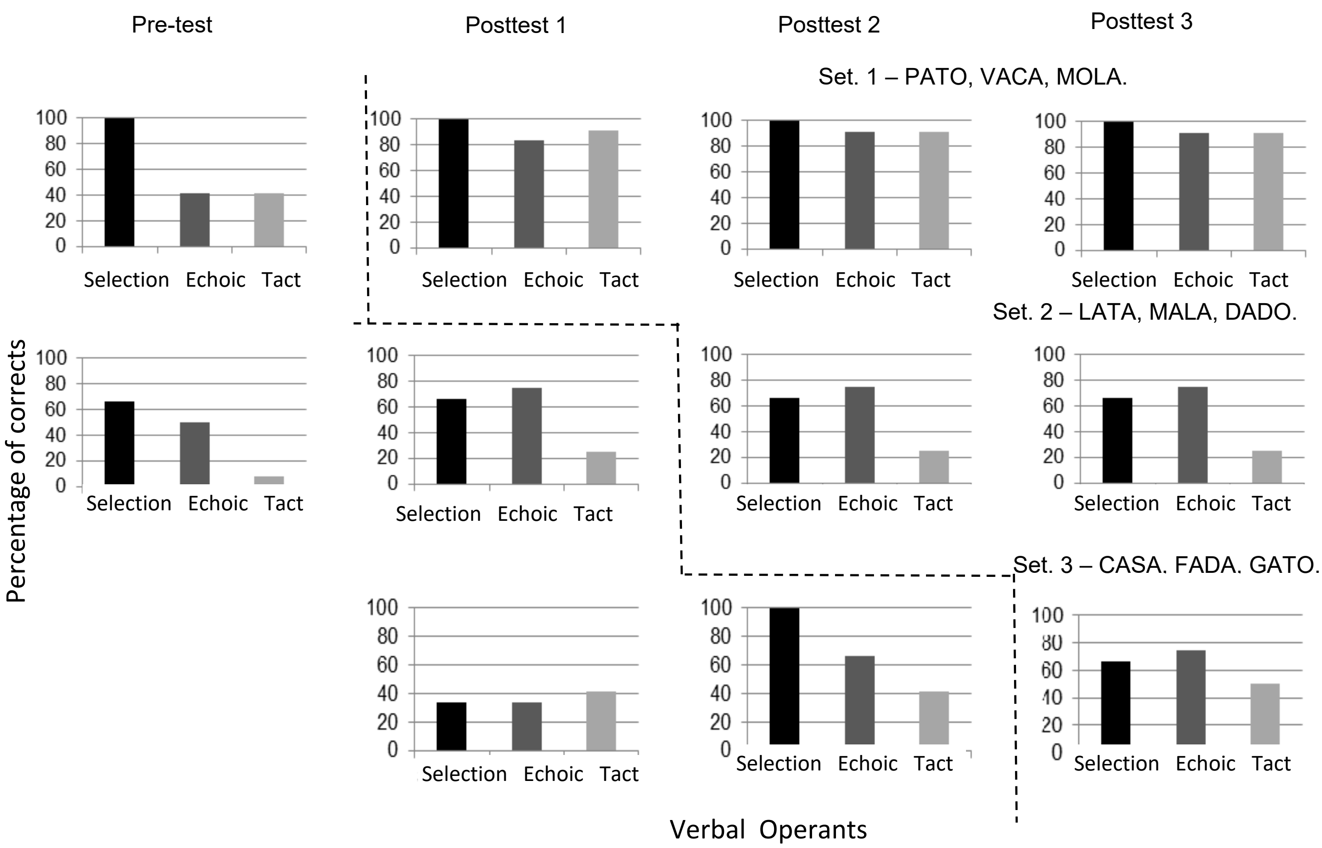


Figure 2. Percentage of correct responses in echoic, selection, and tact operants, after teaching (dotted line) each set of words

After teaching set 2, the participant demonstrated a greater number of correct responses for the selection and tact operants, with a 33.4% increase for the former and 58.3% for the latter, while echoic decreased the number of correct responses by 8.3% for the same stimuli. By measuring the effect of teaching set 2 on performance of sets 1 and 3, we observed that the percentage of correct responses in echoic increased in set 1 by 8.4%, and performance was maintained for the other two operants; in set 3, correct responses increased for selection and echoic, by 66.7% and 33.4% respectively, and was maintained for tact.

After teaching set 3, performance for echoic and tact operants increased by 8.3% for both and selection decreased by 33.4%. Influences of teaching set 1 on set 3 were maintenance of performance for all operants; on set 2 influences were an increase of performance in echoic by 8.3%, and a decrease in the percentage of correct responses by 66.7% in selection and 66.6% for tact.

DISCUSSION

Considering the existing gap in the literature regarding the effectiveness of MEI for populations with developmental delays in language skills, and due to its clinical potential already demonstrated for participants with ASD, the objective of this study was to replicate the results of MEI in the establishment and integration of listener and speaker behaviors, in a child with ANSD. It is possible to state that the procedure was effective in increasing the percentage of correct responses for listener repertoires based on stimulus selection, echoic, and tact responses by making these closer or even superimposed. These results are consistent with the research objectives as well as with other studies involving teaching using MEI[12,13]. This procedure has shown a positive effect on the acquisition of listener and speaker repertoires as well as their integration (functional interdependence) in populations with low verbal repertoire. This enables individuals to behave in social environments, by responding to arbitrary sounds, signs and symbols[13].

Although the participant already exhibited listener repertoire (selection) in acquisition, teaching had the effect of increasing speaker responses with stimuli from sets 1 and 3, and listener and speaker responses with stimuli from set 2 (Figure 1). Listener and speaker behaviors are a prerequisite for learning more complex verbal operants such as, for example, problem solving[12]. In problem solving, the subsequent response may not be the solution, but speaker and listener behaviors interacting in a covert manner, and arriving at the solution response systematically.

The most significant increases in verbal repertoires occurred right after teaching each set (Figure 2, posttests immediately to the right of the dotted line). It is also possible to observe that sets 2 and 3, which were tested twice before being taught, had superior results in the second pre-test. It is possible that teaching set 1, taught the child important skills of integrating listening and speaking, which were then extended to the second pre-test of sets 2 and 3[16].

We observed variability in the participant's repertoire during learning of verbal relations (Figure 1) as well as during pre-tests (Figure 2). Whether this performance is associated to the manifestation of the Auditory Neuropathy Spectrum Disorder (ANSD)[2,25] characterized as an Auditory Dyssynchrony – in other words, as a difficulty in comprehending words in cases of slight to moderate hearing loss, especially in loud environments –, or if other characteristics of the procedure decreased the observed variability and guaranteed response maintenance in successive tests, are empirical questions to be answered by future studies.

On one hand, processing of sound in patients with ANSD is highly variable and fluctuating, and relations between auditory sensibility and capacity to process speech do not follow typical hearing loss rules. Auditory functions may present alterations over time, with progression or variation, or even remain stable[25]. On the other hand, procedures that involve intermittent reinforcement schedules, gradually increasing the quantity of responses emitted for correct or incorrect feedback, may favor maintenance of responses when teaching contingencies become remote.

Children with CI who present fewer deviations in perception and auditory capacity of vocal quality have better perception of speech sounds[26]. Children with ANSD and CI also attain significant improvement in listening and speaking abilities, as shown in several studies[5,27]. Studies report improvement of auditory abilities in 94% of participants with the use of CI, and have shown that electrical stimulation was able to compensate for the characteristic dyssynchrony of ANSD[5]. Even so, the effectiveness of CI is influenced by several variables, such as age, time of use of CI, participation in a systematic therapeutic process and when the diagnosis was made[27].

Although there is research that demonstrates the effectiveness of CI for people with ANSD regarding improvement of auditory performance – as reflected in the bibliographical review, [28] there is no consensus between studies on how and when a child with ANSD presents good development. Currently, research that demonstrates detailed results that compose what is considered improvement auditory performance in children with ANSD is scarce. Listener abilities, such as detection, discrimination, recognition and comprehension, and how listener abilities may interact with speaker[12,13,15] abilities are not specifically detailed.

Considering this gap and the one in research using MEI[29], this study is an important contribution in replicating results obtained with procedures that promote rotation of different types of verbal operants in the integration of listener and speaker repertories. One differential is the population used in the study, because it extends the results obtained most frequently with people with ASD diagnostic characteristics to a child with ANSD. This study contributes to the possibility of planning not only teaching listener and speaker skills to the population with ANSD, but also integration of these repertoires through rotation of stimuli. Thus, the replicability of these results should be verified in future research. On the other hand, the present study also emphasizes several control variables that should be considered in future research, and are further detailed ahead.

The successive posttests (Figure 2) demonstrated the maintenance of results obtained for stimuli from sets 1 (posttests 2 and 3) and 2 (posttest 3) and worked as a follow-up measure. Although maintenance was observed in posttest data from immediately after teaching, the measures obtained were not replicated in the last teaching step (Figure 1). Investigating under which conditions results would be maintained is a question for future studies.

Moreover, as a limitation, this study did not obtain measures of spontaneous use and generalization of words taught in other contexts. New studies could obtain naturalistic measures of taught words, especially when conventional words of common use are selected, as was the case in this study.

Another limitation of this study is the absence of comparisons between objective measures of auditory capacity, such as those obtained by Audiometry, and behavioral measurements, such as a percentage of correct responses in listening tasks based on selection or even performance on scales that characterize auditory abilities of participants (MAIS and MUSS). Future studies could characterize (pre-test) and monitor participants' performance (posttest) by controlling for objective and behavioral measures, observing change curves of repertoires and observing if there is a correspondence between these measures. New research can increase the sample of children with ANSD, thus verifying the replicability of the establishment and integration of listener and speaker repertoires, and isolating extraneous variables that may have influenced the results described in this paper.

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