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Speech pathology telepractice for children with cleft palate in the times of COVID-19 pandemic

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ABSTRACT

Objective: To study whether providing Speech and Language Pathology (SLP) interventions by telepractice (TP) could effectively improve speech performance in children with cleft palate (CCP).

Methods: Forty-three CCP were treated with TP intervention in 45 min sessions, 2 times per week for a period of one month. Children ages ranged 4–12 years (X = 7.04; SD = 2.59). All children presented with velopharyngeal insufficiency (VPI) and compensatory articulation (CA) after palatal repair. TP was provided in small groups (5–6 children) following the principles of the Whole Language Model (WLM). Severity of CA was evaluated by a standardized scale at the onset and at the end of the TP period.

Results: At the onset of the TP intervention period, 84% of the patients demonstrated severe CA. At the end of the TP period there was a significant improvement in severity of CA (p < 0.001).

Conclusion: The results of this study suggests that TP can be a safe and reliable tool for improving CA. Considering that the COVID-19 pandemic will radically modify the delivery of Health Care services in the long term, alternate modes of service delivery should be studied and implemented.

1. Introduction

The ongoing COVID-19 outbreak has rapidly spread through the world. The World Health Organization (WHO) has declared a pandemic and practically every government issued lockdown and quarantine orders.

As a consequence, several measures have had to be implemented severely affecting the entire society at all levels. Everyday activities like school attendance or health care appointments have had to be postponed.

The need to adapt to this unexpected global emergency and the availability of technological tools have enhanced the implementation and expansion of programs aimed to provide options for health care services. Telepractice (TP) is one of such programs. TP is the use of information and communication technologies (ICT's) to deliver health care services where there is physical separation between care providers and/or the recipients over both long and short distances [1].

In the specific case of craniofacial malformations, it is well known that children with cleft palate (CCP) are at risk for a wide range of speech disorders that commonly includes hypernasality, nasal emission, and/or articulation errors such as compensatory articulation (CA) [2]. Hypernasality is excessive nasal resonance and nasal emission refers to escape of air through the nose during articulation of plosive or fricative phonemes. These are the cardinal signs of velopharyngeal insufficiency (VPI). As these disorders are caused by an anatomical deficiency their treatment has to be physical manipulation by surgical procedures or prosthesis. VPI cannot be treated by Speech and Language Pathology (SLP) treatment [3].

In contrast, compensatory errors should be treated by a specific SLP intervention. CA is secondary to VPI and affects speech intelligibility more than VPI with adequate articulation placement. These compensatory errors include dysfunction not only of the velopharyngeal sphincter but the entire vocal tract and frequently require a prolonged period of SLP intervention. Compensatory articulation patterns are a learned articulation deviation. In these cases, the normal articulation placement is substituted by a more posterior placement in the vocal tract [2]. Hence, the child develops atypical placements to meet the pressure-valving requirements for speech. Modifying these patterns is a

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long process. Correcting articulation will improve intelligibility, however, in most cases, hypernasality will persist [2,3]. As stated before, the residual VPI will need to be corrected by a planned surgical procedure and previous imaging studies.

The speech characteristics associated with VPI mentioned herein are the most notorious problems encountered by clinicians treating these children and thus, these disorders have taken a high priority in research and/or intervention for many years [4].

Language disorders in CCP have received relatively little attention in the current related scientific literature [5]. However, a systematic review of the literature revealed that CCP and even adults with cleft palate perform more poorly on cognitive tests than their age-matched peers [4]. These authors also found a high incidence of learning and language disabilities in CCP who had average intelligence. Other studies found similar results; Richman and Nopoulos [6] studied Language performance of CCP in areas such as vocabulary, fluency, rapid verbal labeling, and sentence repetition. They found that CCP present with deficits in both receptive and expressive Language. Other authors found language delays in CCP as compared with non - cleft children [7,8]. However, Other authors [9] reported no significant difference between cleft and non-cleft language development. In sum, the prevalence of language disorders in the cleft palate population is still a controversial issue since there is not a reliable consensus concerning language development in CCP. However, the statement that diagnosis and treatment of communication disorders in CCP should focus not only on speech, but also on higher levels of language processing has been supported by several re-

Different studies [11,12] have reported that SLP treatment in CCP requires a long follow-up. The typical service model of SLP intervention has had to be suspended during the COVID-imposed lockdown. Thus, TP has been implemented in several centers in order to continue with the SLP treatment.

It has been reported that TP enables the improvement of care. Some of the advantages of TP are the following: Facilitates access to specialized medical services, overcomes geographic barriers, and helps in the early identification of health problems, especially in developing countries [13].

The purpose of this paper is to study whether providing SLP intervention by TP could effectively improve speech performance in CCP by correcting articulation patterns involved in CA.

2. Methods

This study was carried out at Hablarte e Integrarte, AC (HeI). HeI is a non-for-profit organization that provides SLP interventions to children of low-income families in Mexico City. HeI is funded with private donations and the treatment costs for the patients are established according to a social service evaluation.

Most of the SLP interventions at HeI are for patients with craniofacial malformations. The Internal Review Board (IRB) of the Institution approved the protocol and the study had been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki's and its later amendments. Before the inclusion of each patient into the study group, the parents or legal guardians were carefully explained the procedures and the methodology of the study. All parents of the patients included in the study group, agreed to participate and gave their informed consent prior to the inclusion of the study.

Sample size was calculated at an Alfa of 95% confidence interval and a Beta power of 80% for a single group study. The distribution of the severity of CA across the patients evaluated in the center during the last 2 years was considered. The aim was to detect an advance greater than 25% as compared to the clinical data of the evaluation at the onset of the intervention. According to these calculations, a minimum of 30 patients should be included in the study group. All CCP who were attending regularly to SLP interventions at the Center before the pandemic were invited to participate in the study. To qualify for the study group,

patients had to meet the following criteria:

- Unilateral, complete cleft of primary and secondary palate (UCLP) [14]. Palatal repair of the UCLP performed according to the same surgical protocol including: Surgical repair of the lip and primary palate between 1 and 3 months and surgical repair of the secondary palate between 6 and 12 months with a minimal incision palatopharyngoplasty [15].
- 2. Absence of postoperative fistulae.
- 3. No known neurological or genetic syndromes.
- 4. Chronological age between 4 and 12 years of age at the time of selection for the study group.
- 5. No identified severe language disorders according to the SDS-Model evaluation practiced in our clinic routinely and reported previously [12].
- 6. VPI after palatal repair demonstrated previously by a complete SLP clinical and perceptual evaluation, videonasopharyngoscopy and multiplanar videofluoroscopy [15].
- 7. CA in association with VPI had to be demonstrated during a complete SP evaluation [12].
- Normal hearing as demonstrated by a previous recent conventional pure-tone audiometry.

2.1. Patients and procedures

All patients already had undergone in-person a complete clinical evaluation of speech, language, resonance and voice. It should be pointed out, that such evaluation is considered as the gold standard diagnostic marker of CA [16,17]. A total of 48 patients and their families agreed to participate and they were selected for the study. Five patients stopped attending the TP intervention (TPI) sessions for different reasons and they were eliminated from the study group.

A Goldman - Fristoe test (Spanish version) of articulation was used for evaluating placement and manner of articulation previously inperson. In addition, for measuring advances in articulation the severity of the CAD was measured according to a previously validated and reported scale. All patients were re-evaluated specifically for articulation placement online, at the onset and at the end of the period of TPI. Special attention was focused on the detection of CA patterns and the phonological rules of the phonological system of each child. For this purpose, children were recorded interacting online with a trained Speech Pathologist during storytelling for 30 min. A 20 min segment was selected where a high level of verbal interaction -occurred. The 20 min of interaction were transcribed verbatim for analyzing the presence and severity of compensatory articulation. Three speech pathologists participated in this study. A blind procedure was utilized for assessing the reliability of the evaluation of CA severity. All analyses were independently conducted by two trained Speech Pathologists with several years of experience treating CPP with VPI and CA. Whenever there was a disagreement, each case was discussed until a consensus was reached. In cases in which there were disagreements or the articulation assessment was not clear a structured phonologically balanced speech sample was used. The articulation of all the children was evaluated according with a previously reported clinical scale of severity of CA [18]. This scale has been used with good results for measuring changes in articulation [18, 19]. It categorizes the degree of severity of CA in each child in 8 levels as follows:

- Constant CA (0). The patient is not able to correct articulation not even in isolated phonemes and despite direct instruction. Intelligibility is severely affected.
- Articulation of isolated phonemes (1). The patient is able to correct articulation only in isolated phonemes through direct instruction. Intelligibility is severely affected.

- Articulation in syllables or words (2). The patient can correct articulation during isolated words, only when the clinician uses specific instruction. Intelligibility is affected.
- Articulation in phrases (3). The patient can correct articulation during phrases with the model of the clinician.
- Articulation within a closed context (4). The patient can articulate
 correctly when talking within a specific context with visual support
 (e.g., when reconstructing a passage from a known storybook using
 visual clues from the illustrations). Nonetheless, he/she shows
 frequent compensatory errors during spontaneous speech, and this
 affect intelligibility.
- Articulation within an open context (5). The patient self-corrects
 articulation when using speech within a specific context. Now he/
 she can connect the information of the storybook and articulate
 correctly while talking about personal experiences. Nonetheless he/
 she shows compensatory errors during spontaneous speech, and this
 affect intelligibility.
- Inconsistent articulation (6). The patient shows compensatory articulation errors inconsistently during spontaneous speech. Intelligibility is not significantly affected
- Appropriate articulation (7). The patient is able to produce adequate placement and manner of articulation during spontaneous speech, including non-present situations.

All patients underwent TPI aimed to correct CA according to the phonologic principles of the Whole Language Model (WLM) [20,21]. All language areas were addressed with emphasis in articulation. The intervention goals were set depending on the phonological needs of each child. TPI was provided by one Speech Pathologist taking care of a small group of CCP (5–6 children). The groups were assembled including children with similar levels of severity of CA according to the evaluation at the onset of the TPI period (not more than 2-level difference), similar ages, similar level of linguistic organization and similar cognitive level.

TPI sessions were imparted for 45 min at a frequency of 1 per week for a period of 1 month (4 weeks, all April). In addition, they were invited to participate in a virtual choir singing session conducted by the Speech Pathologist. Thus, patients were participating in 2 sessions per week, 1 therapy session and 1 choir session. The songs used for the choir singing session included words with plosive and fricative phonemes in combination with high and low vowels. The aim was to practice articulation placement by singing with music. For the music and lyrics a professional musician together with a Speech Pathologist previously prepared these songs. The use of these materials was previously reported [19]. Patients and parents were instructed about strategies and ideas about how to use the materials since the songs were distributed for practicing at home.

The following dependent variable was studied: levels of advance in the severity of CA based on the evaluations at the onset and at the end of the TPI period.

2.2. Telepractice intervention for speech

The TPI was delivered on the ZOOM.US platform. The parents had to previously download the app in their phones or tablets. Some families were able to easily learn how to use the platform. However, for other families the process was not so easy and it took 1-2 sessions for them to be able to smoothly navigate through the sessions. All patients and families included in the study group were able to attend and effectively participate in all sessions during the period of TP.

For TPI the principles of the WLM were followed [20,22,23]. This methodology provides naturally occurring contexts for addressing children's language and articulation and allow for an extended exploration of general concepts. The goal was to work all linguistic areas in an integrated fashion. For CCP, this methodology it's adequate since it approaches both linguistic areas affected in cleft palate, language development and articulation [18,19,21,24]. Thus, articulation goals

were always present as an organized whole. Most activities were conducted as event representations such as storybook reading or singing. Strategies for articulation were used before, during and after each activity. Thus, as the narrative of the storybook or the song was being followed and expanded, the articulation placement, manner and voicing of the target sounds were being reinforced. Both, the storybooks and the songs were carefully selected to meet the needs of each group.

The articulation strategies used during this study have been previously reported and they are regularly used in the center including: Modeling, modeling with stress, cloze procedure with phonemic cues, phonetic changes, and think aloud in phonemic awareness [25]. All strategies were used within structured activities aimed to provide children with contextually appropriate opportunities to include language and focus on articulation. The clinician could choose either to expand an expression or refine upon an expressed idea by giving specific information.

Clinicians also used verbal expansions to provide children with information about higher levels of language organization. This type of interaction has been shown to increase the semantic and syntactic complexity of children's utterances, and may have similar effects upon children's phonological productions [26].

The structure of the session varied depending on the age of the participants but the majority followed the following principles: A storybook was chosen depending on the linguistic level of the group. This gave the context for elaborating the topic of the session. The content and meaning was addressed so that children could become familiar with the language and vocabulary of the story. The topics were related to children's lives such as going to the doctor, to the farm, to visit grandparents, or preparing some food. In this first approach, the speech pathologist modeled articulation and used the strategies described above for engaging participants to comment on the history and practice articulation during the storybook reading. Subsequently, the meaningful words containing the target sounds were practiced with each children in different ways. In most cases, parents were present and participated actively. They were asked to reinforce articulation and to realize the activities with their children.

The age of the patients ranged from 4 to 12 years. Mean age was 7.047, SD = 2.599 with a median of 6.5 years (Table 1).

3. Results

At the onset of the TPI period, 84% of the patients demonstrated severe CA (categories 0 = Constant CA to 3 = Articulation in short phrases as described by the scale of severity).

When comparing levels of severity of CA at the onset and at the end of the study a Two tailed Wilcoxon rank sum test showed a statistically significant difference between the two-paired samples (onset as compared to end). This difference indicates that most patients from the study group demonstrated significant improvement in CA after 1 month of TPI (Table 1).

4. Discussion

Speech therapy for CCP can be a long and difficult path. For children with CA, changing the phonologic rules that have already been established for the production of speech sounds is not an easy task. Different methodologies for treating these disorders have been described. Most of them using a phonetic/articulatory approach [2,3]. Regular attendance to SLP intervention is essential for achieving effective outcomes. Moreover, it has been suggested that the treatment for correcting CA requires intensive stimulation [18]. For many families it can be difficult or even impossible to find accessible speech services near their communities. These difficulties are more frequent and severe in developing countries such as Mexico. CCP present several other risk factors affecting language development. Hoff (2003) reported that children from families with low socio-economic status have poor language skills in a variety of

Table 1Ages and levels of severity of CA of the patients included in the study group.

NO.	AGE (yrs)	CA at Onset	CA at End	LEVELS ADVANCED
1	4	2	4	2
2	4	0	2	2
3	4	3	4	1
4	4	2	3	1
5	4	2	3	1
6	4	0	2	2
7	4	1	3	2
8	4	3	4	1
9	4	3	4	1
10	5	2	5	3
11	5	2	3	1
12	5	2	3	1
13	5	2	4	2
14	5	2	3	1
15	5	2	4	2
16	5	2	4	2
17	5	1	3	2
18	6	3	4	1
19	6	1	2	1
20	6	1	2	1
21	5	2	4	2
22	7	5	6	1
23	7	1	3	2
24	7	4	5	1
25	8	5	6	1
26	8	3	5	2
27	8	0	2	2
28	8	2	4	2
29	8	1	2	1
30	9	3	6	3
31	9	3	4	1
32	9	4	5	1
33	9	4	6	2
34	10	2	4	2
35	10	2	4	2
36	10	4	5	1
37	10	3	4	1
38	10	3	5	2
39	11	4	6	2
40	11	3	5	2
41	11	3	5	2
42	12	1	3	2
43	12	3	4	1

The levels of severity at the onset and at the end of the period of Tele Practice (TP) according to a standardized 8 - category scale are being displayed. Age: Median = 6.5 years; X = 7.047; Range = 4-12; SD = 2.599.

A two - tailed Wilcoxon Rank Sum Test for paired samples demonstrated a statistically significant improvement (p < 0.001) between levels of severity at the onset and at the end of the period of TP.

linguistic domains including vocabulary, grammar, narrative development, phonological development and speed of processing. The proportion of children living in poverty whose language development reflects influence of socioeconomic status is likely to be greater than 22%. Mothers with less education often talk less to their children, make use of a smaller vocabulary and use less complex and varied syntactic structures [27]. All these situations contribute to the vulnerability of the population of CCP in developing countries since they are at a higher risk for presenting with language disorders in addition to the more obvious speech deficits. As a consequence, they require a complete and effective SLP intervention addressing all linguistic areas and skills considering higher levels of linguistic organization such as abstract thought. In this context, phonological information can be integrated with all linguistic areas in all situations with communicative purposes [18-20]. The correct production of speech sounds is no longer the only goal but to guide children to become effective communicators and language users. Thus, the methodology for the SLP intervention used in this study seemed the most adequate for achieving these goals.

In developing countries, for low-income families the situation gets more complicated by not having SLP services available. In these scenarios, TP can be a viable and effective option.

The activities used for the TPI sessions in this study facilitated keeping the children focused. Storybooks and/or songs with video promoted interest and motivation. In this way they are more willing to work and practice articulation. A previous study reported that audiovisual materials could provide an adequate and attractive framework for CCP with CA [19]. These materials were used in the choir sessions and resulted extremely useful for TP. Listening, singing, reading aloud, and role playing are usually attractive activities for children in the age range of the study group studied herein. Linguistic interactions should take place within significant contexts. Other authors have also stated the importance of meaningful contexts for working speech and language [22,28,29]. Also, it has been described that storybook reading is an ideal context for stimulating speech and language [28]. Finally, it is paramount to use the adequate strategies for developing phonologic awareness and promoting articulation correction in CCP and CA during TPI.

The purpose of this paper was to study whether TPI could enhance CA correction in CCP. It was hypothesized that CCP with speech disorders could benefit and improve articulation with TP. The results of this study supported this hypothesis. Children demonstrated improvement in the severity of CA after 1 month of TP. Other studies have found similar results [30]. In a systematic review, when studying whether TP was as effective as conventional in-person delivery for primary school-age children with speech and/or language difficulties, results revealed that both TP and in-person participants made similar and significant improvements. This report concluded that there is limited but promising evidence to support TP for delivering SLP intervention services to school age children. Moreover, the author stated that TP could be an option for rural children where in-person services are limited [31].

Another advantage of TP is that it saves time and costs since families do not have to invest in transportation. Other study supports this statement [32]. In this study, TP was provided to groups of children utilizing real-time videoconferencing facilities. This allowed interacting directly with the participants and to focus and cover individual needs. This finding is consistent with previous studies that indicate that real-time interactions can influence the clinical outcomes since the level of connection enhance the sense of clinician presence and rapport [33]. Moreover, TP is not always an easy task for the clinician. He/she has to develop skills for establishing positive and nurturing interactions in a virtual setting for providing high-quality care to all group members. In addition clear instructions for parents have to be provided before and after the sessions. However, from the results of this study it seems that the use of TP is effective for enhancing the correction of CA in CCP.

The limitations of this clinical trial should not be overlooked. Only a relatively reduced and homogenized number of patients were included in the study group for this paper. The follow up period was short. It should be considered that many patients living in small communities might have difficulties for accessing TP. Although TPI seems to be a promising option, it will be necessary to study larger groups of patients with different levels of severity of CA for longer periods of time and in various locations where attending a formal speech pathology intervention in a clinical or school setting is not feasible. Finally, for this preliminary pilot study no control group was studied in order to compare TP to person-to-person delivery of SLP treatment.

5. Conclusion

Advances in technology have opened the door to countless innovations. The use of TP may be an option for speech pathology treatment. It is probable that COVID-lockdown will contribute to modify the delivery mode of SLP intervention for many clinicians in many instances. This will benefit the large number of families who do not have the opportunity to attend to SLP intervention on a regular basis.

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Declaration of competing interest

All authors declare no conflict of interest.

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