Receptive and expressive language performance in children with and without Cleft Lip and Palate

Desempenho de linguagem receptiva e expressiva em crianças com e sem Fissura Labiopalatina

ABSTRACT

Purpose: To compare the performance in the abilities of receptive and expressive language of children with cleft lip and palate with that of children without cleft lip and palate with typical 12 to 36-month chronological development. Method: The sample consisted of 60 children aged 12 and 36 months: 30 with cleft lip and palate diagnosis and 30 without cleft lip and palate diagnosis with typical development. The groups were paired according to gender, age (in months), and socioeconomic level. The procedures consisted of analysis of medical records, anamnesis with family members, and valuation of the Early Language Milestone Scale (ELMS). The chart analysis showed 63.34% of the children with unilateral cleft lip and palate, 16.66% with bilateral incisive transforamen cleft, and 20% with post-foramen cleft. Children with cleft lip and palate underwent surgeries (lip repair and/or palatoplasty) at the recommended ages and participated in early intervention programs; 40% presented recurrent otitis history, and 50% attended schools. Statistical analysis included the use of the Mann Whitney test with significance level of \( p < 0.05 \). Results: There was a statistically significant difference between the groups regarding receptive and expressive skills. Conclusion: The group of children with cleft lip and palate showed statistically significant low performance in receptive and expressive language compared with children without cleft lip and palate.

RESUMO

Objetivo: Comparar o desempenho nas habilidades de linguagem receptiva e expressiva de crianças com fissura labiopalatina com crianças sem fissura labiopalatina e com desenvolvimento típico de idade cronológica entre 12 e 36 meses. Método: Participaram do estudo 60 crianças, com idade cronológica entre 12 e 36 meses; 30 do grupo experimental, com fissura labiopalatina, e 30 crianças do grupo comparativo, sem fissura labiopalatina, com desenvolvimento típico. Os grupos foram pareados quanto ao gênero, idade cronológica (em meses) e nível socioeconômico. Os procedimentos constaram de análise de prontuário, anamnese com os familiares e da aplicação da Early Language Milestone Scale (ELMS). Na análise do prontuário, verificou-se que 63,34% das crianças apresentavam fissura transforame incisivo unilateral; 16,66%, transforame incisivo bilateral; e 20%, pós-forame. As crianças com fissura labiopalatina realizaram as cirurgias (queiloplastia e/o palatoplastia) nas idades recomendadas e participavam de programas de intervenção fonoaudiológica; 40% apresentaram historico de oitides de repetição e 50% frequentavam escolas. O estudo estatístico contou com a aplicação do teste de Mann Whitney com o nível de significância \( p < 0.05 \). Resultados: Houve diferença estatisticamente significante na comparação entre os grupos em relação às habilidades receptivas e expressivas. Conclusão: O grupo de crianças com fissura labiopalatina apresentou desempenho inferior, estatisticamente significante, nas habilidades de linguagem receptiva e expressiva quando comparado com crianças sem fissura labiopalatina.
INTRODUCTION

Children with complete or incomplete cleft lip and palate (CLP), present higher risk to develop changes in language, however, data in literature are controversial\(^4\). The literature lacks attention on the subject\(^6\).

Several studies along the past twenty years demonstrated that children with CLP show high variability of communication disorders\(^5\) and many focus on speech related issues\(^5-8\). Regarding language performance, there are differences described in the receptive and expressive skills among children with CLP when compared to children without CLP and with regular development, with increased predisposition to delayed acquisition of the first words, sentences production, language comprehension, memory and rationale\(^9-12\).

Studies showed\(^13,14\) that communication changes occur mainly in expressive skills when the CLP is not repaired in appropriate ages, there is dependence of the surgical technique used and when there is the presence of hypernasality and difficulty articulating sounds\(^9,15\).

Regarding receptive language, studies suggest that the presence of early hearing loss could explain the difficulties in the development of language in children with CLP\(^14,15\). Some studies have reported the relationship between hearing loss associated with otitis media with effusion in reducing language scores\(^16,17\).

The early identification and treatment of language disorders in children with CLP are extremely important in order to avoid future emotional problems, socialization, school learning and personal fulfillment\(^13,18-20\).

In this perspective, the analysis of children’s performance in communication and language skills should be checked in the light of intervening factors in the acquisition and development of language.

Therefore, this study aimed to compare the performance in the abilities of receptive and expressive language of children with cleft lip and palate with that of children without cleft lip and palate with typical 12 to 36-month chronological development.

METHOD

This study was approved by the Research Ethics Committee of the Hospital de Reabilitação em Anomalias Craniofaciais of the Universidade de São Paulo (Protocol nº 122/2007-CEP). All ethical criteria were met as regulations of the National Council of Ethics in Research (CONEP), including the signing of an Informed Consent Form by the responsible or guardian of the child.

This cross-sectional study with a sample of 60 children, with ages ranging between 12 and 36 months, divided into two groups: experimental group consisted of 30 children with CLP (GCLP); and comparison group (CG): composed of 30 children without CLP with typical development, matched according to gender, chronological age and socioeconomic status\(^21\).

The following inclusion criteria were considered to select the participants:

GCLP: Present CLP without other associated malformations or genetic syndromes; been submitted to cleft lip and/or palate repair in the recommended ages by international protocol\(^22,23\) used in the Hospital de Reabilitação em Anomalias Craniofaciais; chronological age between 12 and 36 months of age; not born premature and low birth weight; absence of sensorineural hearing loss; absence of visual impairment (low vision).

CG: Absence of CLP, history of typical neurodevelopment; not born premature and low birth weight, absence of sensorineural hearing loss; absence of visual impairment (low vision) and be paired as the chronological age (in months), gender and socioeconomic status.

Procedures

The procedures consisted of: analysis of medical records to collect information such as time of surgical procedures, hospital admissions, results of auditory and visual assessments; anamnesis with the legal representative, including information about the child’s development; Use of the Brazilian Economic Classification Criteria-CCCB\(^23\), which considers the family property (material goods) and member of the education level of head of the family and assessment with the Early Language Milestone Scale - ELMS\(^24\).

ELMS check the performance of children on the following functions: Receptive hearing (AR), Expressive hearing (EA) and Visual (V). The age range considered in the scale varies from zero to 36 months. To start the ELMS application, a vertical line should be drawn in response protocol, exactly the chronological age of the child (in months). All items intersecting this vertical line should be evaluated. The end result is interpreted by manual criteria of this instrument, finding the base and the ceiling. To calculate the base, it retrocedes to the previous items until they are able to three consecutive items of success in each role. The same is done to determine the ceiling, but are tested later items to chronological age until the child fails to three consecutive items. The highest item that the child passes is considered the ceiling. The results were calculated in months for further statistical analysis as instrument manual. The visual function is not presented in this study, since the instrument checks the visual function until the age of 18 months.

Sample characterization

GCLP and CG were composed of 60 children with chronological age between 12 and 36 months (average 24.3 months), 70% male and 30% female. Regarding the socioeconomic classification of the GCLP and CG groups, 23.3% belonged to the lower class; 66.7%, to the upper lower class; and 10%, the lower middle class.

Regarding the type of cleft, 63.34% of children had unilateral cleft lip and palate; 16.66%, bilateral incisive transforaminal; and 20%, post foramen.

All GCLP participants underwent surgical procedures in similar ages, fulfilling the international protocol followed by the hospital, or lip repair at three months of age; primary palatoplasty at 12 months of age, with a variation two months\(^23,24\). Medical records analysis revealed that 40% of GCLP group had otitis.
It is noteworthy that all GCLP individuals and their families participate in early intervention programs and families receive frequent guidance to encourage the child in activities of daily living. All participants in the CG group were enrolled in schools or daycare. In the GCLP group, 50% were enrolled in schools or daycare.

**Statistical analysis**

Statistical analysis included the use of the Mann Whitney test with significance level of p<0.05.

**RESULTS**

Table 1 presents the comparison between the GCLP and CG groups regarding the hearing performance in Receptive and Expressive Hearing functions showing mean scores, standard deviation and results of the statistical test of Mann-Whitney (significance level of p <0.05).

| Variables   | GCLP | CG | GCLP | CG | T value | GCLP | CG | Standard deviation | P-value |
|-------------|------|----|------|----|---------|------|----|--------------------|---------|
| EH          | 22.56| 28.83| 22.5 | 28.23| 2.63    | 6.27 | 6.28| 0.005*             |         |
| RH          | 25.46| 28.23| 26   | 28  | 1.77    | 5.64 | 6.42| 0.027*             |         |

*Significance value p<0.05

**Caption:** EH: Expressive hearing; RH: Receptive hearing; Mann-Whitney test

**DISCUSSION**

Language development is multifactorial and it is necessary to identify factors that interfere in this process, especially in populations considered at risk for changes in language and communication skills, as in individuals with CLP. The literature presents conflicting results regarding language development in children with CLP, specifically regarding the occurrence of receptive problems.

In this study, there was a statistically significant difference when comparing the groups regarding the receptive auditory function. One of the frequently discussed aspects as to receptive skills in individuals with CLP refers to the presence of hearing impairments, as in the case of recurrent otitis that can interfere with the reception of auditory stimuli and understanding messages, as well as bring relevant consequences for the acquisition of language skills. In this sample, 40% had history of otitis. It is noteworthy that the receptive process involves other variables. Receptive skills involve processing information through the development of perceptual processes such as attention, concentration, memory, among others. Hearing care is critical to the development of receptive skills; however, studies on the process of attention in the population with cleft lip and palate are recent and little explored in the specific literature.

Other considerations are necessary. The episodes of ear infections are temporary and, despite the low socioeconomic level of the samples, children participate effectively in intervention and monitoring programs through guidance to families, which can minimize the deleterious effects of these episodes in the long run.

An environment rich in stimuli should be considered in order to develop the auditory reception, encouraging the child to be able to acquire repertoire through life experience, participation in interactive and dialogical events. The study participants with CLP undergo therapeutic procedures from early childhood, which can positively influence their development.

There was a statistically significant difference between the groups regarding receptive and expressive skills. The expressive skills are particularly impaired in children with CLP and are related to changes in articulation and resonance. Studies have shown that changes in joint and hypernasality occur as a result of anatomical malformations, but they can also be observed after the child has been subjected to corrective surgical procedures (lip repair and/or palatoplasty). The expressive skills are related to intrinsic factors (anatomical conditions, genetics, maturity and motivation) and extrinsic factors (environmental influences, surgical technique used in the reconstruction of CLP, among others).

Language acquisition is part of a series of changes in the child’s behavior, marked by the emergence of symbolic behavior and correlated changes in the way they understand and interact with the world, which have an active role in the construction of knowledge. The child acquires knowledge through environmental exploration, manipulation of objects, repeating actions, the domain itself control the body schema and relationships established in situations experienced.

An important aspect that should be considered in the development of a child’s language with CLP is environmental. If the family exposes children to different environments and promotes interaction in different communicative contexts, the child will be able to acquire knowledge of the language more easily. However, one cannot deny the impact of the CLP on how the family deals with this child. A study showed that parents of children with health problems, chronic illnesses or congenital malformations tend to be more permissive and overprotective. The authors showed that in Brazil, parents and family members tend to adopt more protectionist attitudes, different from other cultures that emphasize independence and autonomy values in family relationships.

It is important to note that studies of language development should enhance cultural issues, as these may represent differences in quantifying this variable. In addition to overprotection, periods of hospitalization and reduced opportunities for the child live in different social environments, can also contribute negatively to the communicative development of children with CLP. In this sample, 50% of children were not enrolled in a school.

This study used a screening procedure to check the receptive and expressive skills of children with CLP. This instrument allows the child control in periodic reviews, which, added to the intervention procedures and frequent family guidelines favor the longitudinal monitoring of the performance of children. Regular application of this instrument allows you to check the child’s performance over time and favors the team of professionals to evaluate the efficacy of therapeutic planning for these skills.
The longitudinal follow-up of these children and language development is essential.

CONCLUSION

The group of children with cleft lip and palate presented a lower performance, statistically significant, in receptive and expressive language skills compared to children without cleft lip and palate with typical development of chronological age between 12 and 36 months.

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Author contributions

DACL was responsible for the study design, data collection, analysis and manuscript writing; MJFSM was responsible for data collection and analysis; CCR was responsible for data collection and analysis; LPM was responsible for study design, data collection, analysis and manuscript writing.