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# Symptoms of dysphagia in children with cleft lip and/or palate pre- and post-surgical correction

## *Sintomas de disfagia em crianças com fissura labial e/ou palatina pré e pós-correção cirúrgica*

### Keywords

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### Descritores

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### ABSTRACT

**Purpose:** Determine the occurrence of dysphagia symptoms in children with cleft lip and/or palate (CLP) pre- and post-surgical correction. **Methods:** Quantitative observational cross-sectional study. Existence of cleft lip and/or palate without association with other syndromes was the study inclusion and/or exclusion criterion. Parents and/or legal guardians responded to a recall questionnaire on the identification of occurrence of coughing, choking, vomiting, and nasal escape pre- and postoperatively and whether these symptoms disappeared after surgical correction. The study was approved by the Research Ethics Committee of the aforementioned Institution under protocol no. 1573164. **Results:** The sample comprised 23 children with mean age of 48 months, mostly male and with unilateral trans-foramen incisor clefts. Statistically significant difference was observed between the pre- and post-surgical periods regarding the presence of dysphagia symptoms. **Conclusion:** Surgical treatment of patients with cleft lip and/or palate proved to be a resource to prevent the occurrence of dysphagia symptoms when associated with adequate intervention chronology.

### RESUMO

**Objetivo:** Verificar a ocorrência dos sintomas de disfagia em crianças com fissura labial e/ou palatina pré e pós-correção cirúrgica. **Método:** Trata-se de um estudo observacional do tipo transversal, de caráter quantitativo. Os critérios de inclusão e/ou exclusão foram de portadores de fissura labial e/ou palatina, sem outras síndromes associadas. Os responsáveis responderam a um questionário com questões de caráter recordatório quanto à identificação de ocorrência dos sinais e sintomas: tosse, engasgo, vômito e escape nasal no momento pré-correção cirúrgica, e o seu desaparecimento ou não, pós-correção cirúrgica. Este estudo foi aprovado pelo Comitê de Ética em Pesquisa sob o protocolo número 1573164. **Resultados:** Amostra composta por 23 crianças com idade mediana de 48 meses, sendo a maioria do gênero masculino e portadora de fissura transforame incisivo unilateral. Houve diferença estatística da presença de sintomas de disfagia entre o momento pré e pós-cirúrgico. **Conclusão:** A correção cirúrgica dos portadores de FLP se mostrou um recurso de prevenção da ocorrência dos sintomas de disfagia, quando associada ao tempo adequado da cronologia de intervenção.

Study carried out at Ambulatórios de Especialidades, Sistema Único de Saúde – SUS, Hospital da Criança Santo Antônio – HCSA, Irmandade Santa Casa de Misericórdia de Porto Alegre – ISCMPOA, Porto Alegre (RS), Brasil and at the Departamento de Fonoaudiologia, Universidade Federal de Ciências da Saúde de Porto Alegre – UFCSPA, Porto Alegre (RS), Brasil.

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## INTRODUCTION

Cleft lip and/or palate (CLP) are among the main congenital craniofacial malformations. They occur because of a fusion error in the embryonic facial processes, more precisely in the gill or pharyngeal processes, between the 4<sup>th</sup> and the 9<sup>th</sup> weeks of embryonic life<sup>(1)</sup>. These anomalies are considered common, presenting prevalence of 1/1000 births in Brazil. Their etiology is multifactorial and may be associated with factors such as heredity, maternal aspects, stress, infections, medication, and irradiation<sup>(1,2)</sup>.

In the Brazilian specific scientific literature, the reference point for classifying clefts of the lip and/or palate is the borderline structure between the primary and secondary palates, that is, the incisor foramen<sup>(3)</sup>. According to this classification, clefts are divided into four categories: pre-foramen incisor, which involves the lip, alveolar ridge, and premaxilla, and may be unilateral, bilateral or median, and complete or incomplete; post-foramen incisor, which affects the uvula and palate, totally or partially, and may complete or incomplete, but generally median; trans-foramen incisor, which comprises the lip, alveolar ridge, and the entire palate, unilaterally or bilaterally; and rare clefts of the face, which involve the lower lip or the nose, and may be oblique or transverse<sup>(3)</sup>.

Surgical correction of CLP is possible, presenting different chronology and techniques depending on the type and extent of the cleft, and the deciding factor is at the surgeon's discretion. The following procedures can be performed for CLP correction: cheiloplasty, lip repair; staphylorrhaphy, soft palate repair; and palatoplasty, hard palate repair<sup>(2)</sup>.

CLP patients present functional difficulties in the mechanisms of swallowing, sucking, chewing, and phonation because of anatomical alterations of the stomatognathic system<sup>(4)</sup>.

Deglutition is defined as the process of conducting food from the mouth to the stomach, and is divided into three main phases: oral, pharyngeal, and esophageal; it occurs voluntarily in the first phase and involuntarily in the latter two phases. The whole swallowing process involves neuromuscular participation of the stomatognathic system structures<sup>(4,5)</sup>.

Dysphagia is defined as difficulty in swallowing, and is characterized as a disorder in face of problems in the transport of food or saliva from the mouth to the stomach, and it may occur at any stage of the swallowing process<sup>(5-7)</sup>.

For safe deglutition, it is necessary that the structures involved be intact, that a correct synchronization between their phases occur, that there be no stasis - presence of food in the oral and/or pharyngeal cavity, and that there be correct glottic closure for protection of the airways, preventing food from escaping into the lower respiratory tract<sup>(5,7,8)</sup>.

Classification of dysphagia impairment in pediatrics, using an assessment protocol, is as follows: normal swallowing, mild oropharyngeal dysphagia, moderate to severe oropharyngeal dysphagia, and severe oropharyngeal dysphagia<sup>(9)</sup>. The most common symptoms of dysphagia are anterior escape; premature posterior food escape; reduction of oral sensitivity; delayed onset of swallowing; laryngeal penetration or laryngotracheal aspiration, generating signs such as coughing, choking, and vomiting during or after eating<sup>(9,10)</sup>. Dysphagic children may present delayed development of oral motor functions, chronic

respiratory diseases, gastroesophageal reflux, weight loss, malnutrition, and selectivity of foods and consistencies<sup>(11)</sup>.

Studies have reported that the main complaints of mothers about the initial feeding process of children with CLP are nasal reflux, choking, coughing, and difficulties in the sucking process<sup>(8,10)</sup>.

In cases of CLP, there are difficulties in establishing efficient and safe suction due to weak intraoral pressure during the performance of this function, as well as to disorders in the suction/deglutition/respiration coordination. An extreme anatomical alteration of the palatal portion of the premaxilla opposes to the movement of the tongue, which hinders the grasp of the breast and/or nursing bottle nipple, which together with the posterior position of the tongue at rest hampers the muscular impulse during the grasp, and/or because of undesirable coupling between the oral and nasal cavities, factors that accentuate food process impairment<sup>(8)</sup>.

Changes in the oropharyngeal structures caused by CLP are considered risk factors for the development of dysphagia. The degree and characteristics of eating difficulties in this population are directly associated with the extent and type of the cleft<sup>(5,10,11)</sup>.

The objective of this study was to determine the occurrence of dysphagia symptoms in children with cleft lip and/or palate pre- and postoperatively.

## METHODS

This qualitative, observational, cross-sectional study was approved by the Research Ethics Committee of the aforementioned Institution under protocol no. 1573164/2016.

Existence of cleft lip and/or palate (CLP) without association with other syndromes in children cared at the Speech-language Pathology Service of a specialty clinic of the Brazilian Unified Health System (SUS) at a pediatric hospital in Porto Alegre, Rio Grande do Sul state, Brazil was the study inclusion and/or exclusion criterion. These children were assisted by six students of the Speech-Language Pathology course of the Universidade Federal de Ciências da Saúde de Porto Alegre – UFCSPA, who belonged to an extension project entitled “Assistance to CLP Patients”, supervised by three professors of the Department of Speech, Language and Hearing Sciences of this institution.

The parents and/or legal guardians of the children were invited by the assistant researcher to participate in the study while they were awaiting consultation at the outpatient clinic or by telephone with a request for an appointment at their convenience. All participants signed an Informed Consent Form (ICF) prior to study commencement.

A questionnaire prepared by the researchers was read by the assistant researcher and answered by the patients' parents and/or legal guardians. The questionnaire aimed to obtain general information (age, date of birth, and gender) on the child and parent, family history of possible heredity, and classification of the cleft. It included closed and open recall questions on the pregnancy, corrective surgeries performed, feeding route at birth, length of time of this feeding route, information received by the parents from professionals regarding posture and general information about feeding, and age of introduction of other food consistencies (pasty and solid). The questionnaire also included identification of occurrence of coughing, choking, vomiting, and nasal escape in the liquid, pasty and solid consistencies.

Finally, it requested information on comparison of the dysphagia symptoms pre- and postoperatively and whether they had disappeared after surgical correction.

The data obtained were stored in a Microsoft Excel® 2010 datasheet and statistical analysis of sample distribution was performed using the Statistical Package for the Social Sciences (SPSS) 16.0 for Windows®. The quantitative variables were described as mean and standard deviation or interquartile range and median, whereas the categorical variables were expressed as absolute and relative frequencies. The McNemar's Chi-squared test was used to compare the symptoms of dysphagia pre- and post-surgical correction. The Fisher's exact test was applied to correlate the dysphagia symptoms and type of cleft. A significance level of 5% ( $p \leq 0.005$ ) was adopted for all statistical analyses.

## RESULTS

The study sample was composed of 23 children, 69.6% male and 30.4% female, aged 15 to 132 months, mean age of 48 months. Regarding the family history of heredity, five (21.7%) reported having case(s) of cleft lip and/or palate (CLP) in the family and 18 (78.3%) did not. The types of CLP and number of cases found were as follows: unilateral trans-foramen incisor, 13 (56.5%); bilateral trans-foramen incisor, three (13.0%); incomplete pre-foramen incisor, two (8.7%); incomplete pre-foramen incisor, one (4.3%); incomplete post-foramen incisor, four (17.4%).

Of the 23 children, two had not undergone any surgical correction until the moment of application of the questionnaire and one had undergone only cheiloplasty and was waiting to schedule a palatoplasty. Table 1 shows the chronology of surgical correction of the study sample.

At birth, the study participants presented the following routes of feeding: 13 (56.5%), nursing bottle; six (26.1%), maternal breast; four (17.4%), feeding tube. Graph 1 depicts the length of time of the initial feeding route.

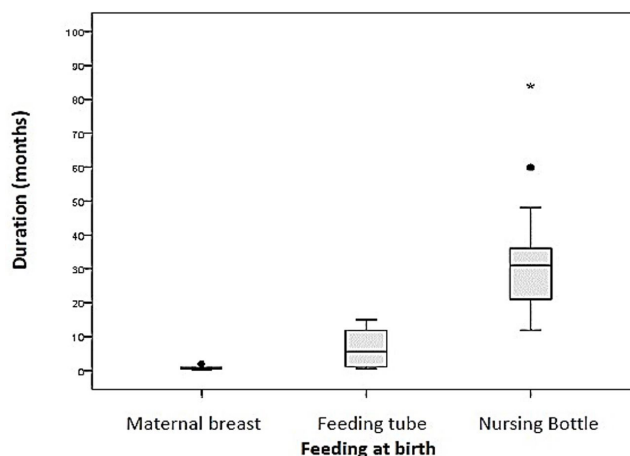
Regarding the mean length of feeding time, 14 (60.9%) reported feeding time  $\leq 30$  min and nine (39.1%) reported time  $> 30$  min. The parents and/or legal guardians of the study participants confirmed whether they had received or not information concerning the feeding process, and informed from which professional and at what moment such guidance was provided. These data are presented in Table 2.

Table 3 shows the proportion of occurrence of dysphagia symptoms according to the type of food offered. At the time of study completion, 18 (78.3%) of the children preferred foods of solid consistency and five (21.7%) of pasty consistency.

Statistically significant difference was observed in the correlation between the symptoms of dysphagia pre- and postoperatively to palatoplasty ( $p \leq 0.005$ ). This correlation is shown in Table 4.

**Table 1.** Chronology of surgical correction

Surgical Variables	n (%)
<b>Cheiloplasty</b>	<b>16</b>
0-3 Months	4 (25.0%)
4-6 Months	3 (18.8%)
7-12 Months	5 (31.3%)
>12 Months	4 (25.0%)
<b>Palatoplasty</b>	<b>17</b>
Up to 18 Months	12 (70.6%)
19-24 Months	3 (17.6%)
>24 Months	2 (11.8%)



\*discrepant value 2.5 above the standard deviation; •discrepant value 1.5 above the standard deviation

**Graph 1.** Length of time of feeding route

**Table 2.** Feeding process information

Variables	n (%)
Has received information about posture and general feeding	n=23
Yes	18 (78.3%)
No	5 (21.7%)
Informing professional	n=18
Speech-language pathologist	8 (44.4%)
Physician	9 (50.0%)
Others	1 (5.6%)
When the information was received	
During the term of pregnancy	0 (0.0%)
At the maternity hospital	9 (50.0%)
During the postpartum follow-up	5 (27.8%)
After clinical care	4 (22.2%)

**Table 3.** Proportion of occurrence of dysphagia symptoms according to the type of food offered

Variables	n (%)
<b>Symptoms with liquid consistency</b>	<b>n (%)</b>
Choking	13 (56.5%)
Coughing	12 (52.2%)
Vomiting	10 (43.5%)
Nasal escape	21 (91.3%)
<b>Symptoms with pasty consistency</b>	<b>n (%)</b>
Choking	8 (34.8%)
Coughing	9 (39.1%)
Vomiting	4 (17.4%)
Nasal escape	20 (87.0%)
<b>Symptoms with solid consistency</b>	<b>n (%)</b>
Choking	9 (39.1%)
Coughing	9 (39.1%)
Vomiting	4 (17.4%)
Nasal escape	16 (69.6%)

**Table 4.** Comparison between dysphagia symptoms before and after palatoplasty

Variables	Before (n=23)	After (n=20)	<i>p</i> *
Nasal escape	22 (95.7)	2 (8.7)	<0.001
Choking	13 (56.5)	1 (5.0)	0.002
Coughing	14 (60.9)	0 (0.0)	0.001
Vomiting	10 (43.5)	1 (5.0)	0.004

\*McNemar's test

Of the 20 participants who had undergone surgical correction, one remained with nasal escape, one with vomiting, and one with nasal escape and choking; all with unilateral trans-foramen incisor cleft.

It is worth mentioning that the three individuals who had not undergone surgical correction remained with presence of dysphagia symptoms.

No statistically significant correlation ( $p \geq 0.005$ ) was observed between the symptoms of dysphagia and type of cleft, but participants with unilateral and bilateral trans-foramen incisor cleft presented higher occurrence of symptoms. Table 5 presents a comparison between the signs and symptoms of dysphagia according to type of cleft.

**Table 5.** Comparison of signs and symptoms of dysphagia according to type of cleft

Signs and Symptoms of Dysphagia	Unilateral trans-foramen incisor (n=13) n (%)	Bilateral trans-foramen incisor (n=3) n (%)	Incomplete pre-foramen incisor (n=2) n (%)	Complete post-foramen incisor (n=1) n (%)	Incomplete post-foramen incisor (n=4) n (%)	<i>p</i> *
<b>Before</b>						
Nasal escape	13 (100)	3 (100)	1 (50)	1 (100)	4 (100)	0.127
Choking	5 (38.5)	2 (66.7)	2 (100)	1 (100)	3 (75.0)	0.318
Coughing	5 (38.5)	3 (100)	1 (50)	1 (100)	4 (100)	0.053
Vomiting	7 (53.8)	0 (0.0)	2 (100)	0 (0.0)	1 (25.0)	0.408
<b>After</b>						
Nasal escape	2 (15.4)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1.000
Choking	1 (7.7)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1.000
Coughing	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	-
Vomiting	1 (7.7)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1.000

\*Monte Carlo simulation for Fisher's exact test

## DISCUSSION

Heredity is among the relevant risk factors for occurrence of craniofacial malformations<sup>(12)</sup>. In this study, the proportion of heredity observed was lower than that reported in the literature, perhaps owing to the reduced sample size. Prevalence of the male gender with unilateral trans-foramen incisor cleft is also described in published epidemiological studies, considering its greater incidence compared with the female gender<sup>(12,13)</sup>, with the same also occurring with respect to earlier fusing time of the nasal and palatal processes<sup>(13)</sup>.

Regarding cleft lip and/or palate (CLP) surgical correction, despite the different chronology described in the literature<sup>(14-16)</sup>, most Speech-language Pathology services recommend that the surgery be performed during the postnatal period, with ideal time for cheiloplasty at three or between three and six months of age, depending on the clinical conditions necessary to perform a risk procedure<sup>(2,17)</sup>.

As for palatoplasty, studies state that it should be performed at twelve or between twelve and eighteen months of age. In cases of complete cleft palate, research suggests posterior palatoplasty, or staphylorrhaphy together with cheiloplasty or palatoplasty, or yet that the procedures should be performed only at eighteen months of age<sup>(2,17)</sup>.

The surgical chronology to which most of the participants in this study was submitted suggests a delay in the performance of cheiloplasty in relation to the ideal time, and more frequent than that found in the literature for palatoplasty<sup>(17)</sup>. The ideal time for correction of both cleft lip and cleft palate should consider the patient's age and functional and individual factors such as the associated anatomical compromises, seeking the best functional and aesthetic repair<sup>(18)</sup>.

The different moments of surgical correction observed in this study are due to the difficulties in maintaining a schedule for these interventions, as they demand stability of the child's health and availability of beds and operating rooms at the Brazilian Unified Health System (SUS). Late correction slows

the rehabilitation of impairments in the sucking, chewing, swallowing and speech functions.

Newborns feed through suction, which is coordinated with breathing and swallowing. Initially, this function is a reflex and is elicited by the touch of the nipple on the infant's lips, which open and suck the nipple, initiating the suction movements<sup>(8,19)</sup>.

At birth, it is recommended that babies be fed exclusively on breast milk, until the age of six months. Breast milk is a complete food, important for immunization and prevention of infections and allergies, and it also assists with the process of bone growth and development of the craniofacial musculature<sup>(20)</sup>. Although with greater difficulty, infants with CLP are able to succeed in the process of exclusive breastfeeding<sup>(6,21)</sup>, considering adequate positioning and adjustments to breastfeeding intervals<sup>(8)</sup>.

The low number of participants in this study who were breast fed, as well as the short length of time in this feeding route, are in agreement with findings in the literature<sup>(4,10)</sup>, and they reinforce the importance of the presence of trained professionals in care teams at birth.

In this study, the nursing bottle was the most commonly observed feeding route, and the one with the longest length of time. The high proportion of nursing bottle as a feeding route in post-birth CLP is also described in other studies<sup>(4,10,22)</sup>. The use of this utensil is considered a deleterious oral habit by the population in general, varying according to its frequency and way of use<sup>(23,24)</sup>.

In the cases of CLP, the nursing bottle, even with no consensus in the literature, can assist with the feeding process because of the existence of nipples shaped to facilitate the grasp and the type of hole suitable for the flow of milk, assisting with the sealing of the structures and favoring the necessary pressure for suction<sup>(4,6,17)</sup>.

Indication of the feeding tube as a feeding route for infants with CLP is necessary only in cases of impossibility of oral feeding, associated with difficulties in gaining weight<sup>(25)</sup>. In this study, 17.4% of the sample used a feeding tube, with length of time of approximately six months. A study that analyzed the medical records of 137 patients with CLP observed that 23% of the sample used a feeding tube; a percentage higher than that found in the present study<sup>(25)</sup>.

Effective suction in infants with CLP varies according to the discontinuity or not of the lips. Because of these factors, this function may be poorly conducted, being associated with modification of intraoral pressure, which can directly affect its effectiveness and, hence, the feeding duration<sup>(6,8,22)</sup>. In this study, predominance of the reported mean length of feeding time was  $\leq 30$  min, corroborating the literature, which describes longer feeding duration for CLP patients<sup>(6,17)</sup> compared with that of the healthy child population; studies indicate that the mean length of feeding time of healthy infants is approximately 20 min<sup>(17)</sup>.

Accuracy of the feeding duration, approximately 30 minutes, reported in this study can be questioned because of the recall method adopted, considering that it can be influenced by the long-term memory of the parents and/or guardians. Most of the parents participating in this research received guidance on posture and general information regarding feeding at some time during hospitalization or in the neonatal period. The literature presents a study in which 26% of the mothers reported receiving guidance on feeding<sup>(22)</sup>, a proportion lower than that found in this survey.

Information regarding positioning of the infants with CLP and their feeding was predominantly provided by physicians and speech-language therapists, and 50% of the participants reported having received this information during the stay at the maternity hospital. It is important for parents and/or caregivers of children with CLP to be informed by health professionals about the ideal posture for feeding and general care for safe feeding<sup>(22)</sup> in view of the possibility of deglutition disorders.

After six months of life, infants need more nutrients to develop; the introduction of new foods is necessary to meet the new nutritional demand. Late introduction of food can cause weight loss and deficit of nutrients that are important for growth<sup>(26)</sup>.

The time of introduction of pasty and solid foods observed in this study was adequate, in agreement with the ages suggested and described in the literature<sup>(26,27)</sup>. At the time of study completion, the preference of the participants for solid food suggests that the process of food introduction occurred properly as expected.

Occurrence of feeding difficulties in CLP patients is widely described in the literature<sup>(4,6,8,10,20-22)</sup>. These difficulties arise at birth, because of impairment of the functions of suction and swallowing<sup>(26,27)</sup>.

CLP has been the focus of different clinical and epidemiological trials and case reports, but few have associated this anomaly with dysphagia so far.

Presence of nasal escape in the different consistencies was the symptom of dysphagia most frequently referred by the study participants. This finding can be justified by the larger number of trans- and post-foramen incisor clefts observed owing to presence of anatomical and structural alterations of the lip, alveolus, and hard and soft palates, which favor food reflux into the nasal cavity.

No statistically significant correlation was found between the symptoms of dysphagia and type of cleft; however, the literature reports data of higher occurrence of feeding problems in trans- and/or post-foramen incisor clefts, in agreement with the findings of the present study<sup>(6,8,28)</sup>.

As described in the literature<sup>(9,28)</sup>, occurrence of coughing, gagging, and vomiting are considered symptoms of dysphagia. Even the term dysphagia is not usually used to refer to eating difficulties in CLP. The findings of this study demonstrate presence of dysphagia in CLP patients pre- and postoperatively.

This study statistically demonstrated the non-prevalence of dysphagia symptoms in the comparison between the pre- and post-surgical correction periods, considering that the symptoms are associated with anatomical deformity of the structures of the stomatognathic system<sup>(8,10)</sup>.

The postoperative occurrence of dysphagia symptoms found in this study, namely, nasal escape, choking, and vomiting, may be associated with the presence of fistulas, from velopharyngeal incompetence and/or insufficiency<sup>(8,17,29)</sup>. The palatine fistula is a failure in the surgical closure of the palate<sup>(28)</sup>, and it can be corrected by a new surgical intervention. Velopharyngeal incompetence is caused by decreased velar mobility and insufficiency due to anatomical or iatrogenic changes, which prevent the pharyngeal sphincter from closing<sup>(8,17)</sup>.

Occurrence of dysphagia in patients with CLP, verified since the neonatal period, interferes with the children's quality of life and feeding. The absence of specialized monitoring and timely surgical correction compromises the overall health of these children and may trigger nutritional, hydration, and general developmental disorders.

When the interventions occur in due time and monitoring has adequate family involvement, they contribute to the better quality of life and development of children with CLP.

It is suggested that further studies on the theme be conducted with larger samples, because the process of correction and rehabilitation of CLP accompanies child growth and development and involves, at least, multiprofessional action, with monitoring of professionals from the areas of Speech-language Therapy, Orthodontics, Oral and Maxillofacial Surgery, Plastic and Aesthetic Surgery, as well as of other sciences.

It is important that surgical correction be performed at the appropriate time, because dysphagia and eating problems may interfere with the quality of life and development of children with CLP. Restructuring of facial deformity and its evolution involve the life cycles of childhood and adolescence. Speech-language pathology monitoring of this population, from birth, aims at assisting with family guidance and therapeutic clinical support to children throughout their development.

## CONCLUSION

Surgical treatment of patients with cleft lip and/or palate proved to be a resource to reduce the prevalence of dysphagia symptoms.

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### Author contributions

*JSF was responsible for the study design, collection and analysis of the data, and preparation and writing of the manuscript; MCAF was the study adviser, participated in its design and critical analysis, data analysis, and preparation and writing of the manuscript.*