



Study of the prevalence of morphological varieties of cleft lip and palate in a tertiary medical center

Estudo da prevalência das variedades morfológicas de fissuras labiopalatinas em um centro médico terciário

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■ ABSTRACT

Introduction: To verify the prevalence, in a specialized medical center, of the different morphological varieties of orofacial clefts and classify them with details of the affected anatomical structures. **Methods:** Observational, cross-sectional, retrospective study, based on a review of medical records of children born between 1989 and 2014 who had some kind of cleft lip and/or palate. The type of cleft was analyzed regarding the incisive foramen. Besides, details on the involvement of the lip, dental arch, nasal filter, soft palate, and/or hard palate and uvula were also collected. **Results:** 1078 medical records of patients with cleft lip and palate were analyzed. When comparing genders, in females, there was a predominance of post-foramen cleft (16.7%), while in males, trans-foramen was more prevalent (25.2%). Separating the cleft as unilateral or bilateral, unilateral ones appeared in more than 75% of patients. Regarding the pre, post, and trans-foramen position, the first two appeared similarly, about 30% each, while the trans-foramen appeared in about 40%. As for the side, the unilateral slits on the left were predominant in both sexes (62% of the unilateral ones). Regarding the anatomical details of the structures involved, the most common injuries were those that involved from the lip to the soft palate. **Conclusion:** The most prevalent type of cleft was unilateral trans-foramen. In the female gender, the post-foramen cleft, and in the male, the transformation was the most frequent. Unilateral fissures were more common than bilateral fissures, and when unilateral, there was a predominance of lesions on the left.

Keywords: Cleft Palate; Child; Cleft lip; Craniofacial abnormalities; Surgery, Plastic.

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■ RESUMO

Introdução: Verificar a prevalência, num centro médico especializado, das diferentes variedades morfológicas de fissuras orofaciais e classificá-las com detalhes das estruturas anatômicas acometidas. **Métodos:** Estudo observacional, transversal, retrospectivo, baseado em revisão de prontuários de crianças nascidas entre os anos de 1989 e 2014, que apresentassem algum tipo de fissura de lábio e/ou palato. Foi analisado o tipo de fissura, tendo por referência o forame incisivo. Além disso, detalhes sobre o acometimento do lábio, arcada dentária, filtro nasal, palato mole e/ou palato duro e úvula também foram coletados. **Resultados:** Foram analisados 1078 prontuários de pacientes com fissura lábio palatina. Comparando entre os sexos, no feminino houve predomínio da fissura pós-forame (16,7%), enquanto que no masculino a transforame foi mais prevalente (25,2%). Separando as fendas como uni ou bilaterais, as unilaterais apareceram em mais de 75% dos pacientes. Em relação a posição pré, pós e transforame, as duas primeiras apareceram de forma semelhante, cerca de 30% cada, enquanto a transforame apareceu em cerca de 40%. Quanto ao lado, as fendas unilaterais à esquerda tiveram predomínio em ambos os sexos (62% das unilaterais). Sobre o detalhamento anatômico das estruturas envolvidas, as lesões mais comuns foram aquelas que envolveram desde o lábio até o palato mole. **Conclusão:** O tipo de fissura mais prevalente foi a transforame unilateral. No sexo feminino a fissura pós-forame e no masculino a transforame foram as de maior ocorrência. As fissuras unilaterais foram mais comuns do que as bilaterais e quando unilaterais houve um predomínio de lesões à esquerda.

Descritores: Fissura palatina; Criança; Fenda labial; Anormalidades craniofaciais; Cirurgia plástica.

INTRODUCTION

Palate development occurs between the fifth and twelfth weeks of embryonic life, with the most critical period occurring between the sixth and ninth weeks. Cleft lip and palate (CLP) occur due to the non-fusion of the maxillary and nasomedial processes during the embryonic period¹.

Among craniofacial malformations, CLP are the most common. In a large study conducted in several Latin American countries, the prevalence of this malformation was 0.87 per thousand births when the cleft lip is evaluated with or without the palate's involvement and 0.13 per thousand births when exclusively cleft palate is evaluated².

Several etiological factors are related to CLP, including genetic, environmental factors, and exposure to teratogens. Thus, malformations are essentially multifactorial and not yet fully defined¹. CLP are generally classified according to the shape and extent of the structures involved. One of the most commonly used classifications is the Spina classification, which is

based on the incisive foramen³. According to the degree of involvement, this classification is subdivided into complete and incomplete, uni, or bilateral.

OBJECTIVE

Thus, this study's objective was to verify the prevalence, in a specialized care center, of the different morphological varieties of CLP based on the Spina classification, in addition to classifying them by the details of the affected structures.

METHODS

It is an observational, cross-sectional, retrospective study based on a review of medical records of individuals born between 1989 and 2014 and followed up at a tertiary medical center specialized in treating craniofacial malformations and located in the south of Brazil. The institutional ethics committee approved the research under number 1,779,572 in 2016. The variables analyzed were the patient's

gender and the type of cleft, based on Spina's model. The fissures were divided into (with due variations in laterality, right, left, or median): incomplete unilateral pre-foramen fissure, incomplete bilateral pre-foramen fissure, complete unilateral pre-foramen fissure, complete bilateral pre-foramen fissure, unilateral trans-foramen fissure, bilateral trans-foramen fissure, incomplete post-foramen fissure, complete post-foramen fissure, rare fissure. Besides, details on the involvement of the lip, dental arch, nasal filter, soft palate, and/or hard palate and uvula were also collected. The information collected was tabulated in Excel spreadsheets and submitted to statistical analysis. Pearson's chi-square and Fisher's exact tests were used to assessing possible correlations between variables. Statistical analysis was performed using the Statistical Package for Social Science (SPSS) program. The level of significance considered was 0.05.

RESULTS

There were found 1,151 cases registered at the medical center between 1989 and 2014. The medical records of individuals with other types of facial malformations were excluded and those with incomplete or missing information, in a total of 73. Thus, the final sample was 1,078 individuals.

Regarding the sex of registered cases, male patients are more common (585 - 54.3%) than female patients (493 - 45.7%) ($p = 0.005$). The cleft with the greatest presence in females was the post-foramen type with 180 cases (16.7%), unlike the male sex, which had a greater number of patients with cleft foramen with 272 cases (25.2%) (Table 1).

Table 1. Absolute frequency (N) and proportion, in percentage, of cleft lip and/or palate location found concerning sex.

Fissure type	Female		Male		Total	
	N	%	N	%	N	%
Post-foramen	180	16.7	132	12.2	312	28.9
Pre-foramen	146	13.5	181	16.8	327	30.3
Trans-foramen	167	15.5	272	25.2	439	40.7
Total	493	45.7	585	54.3	1078	100

Comparing the clefts as to their unilateral or bilateral presence, in those that can be classified as such, that is, disregarding the post-foramen clefts, it was observed that, of a total of 766 cases, 591 (77.2%) are unilateral and 175 (22.8%) are bilateral, regardless of gender, approximately three times more prevalent than unilateral than bilateral.

Unilateral clefts can occur on the right or left side. The distribution of unilateral clefts located on the

left was 368 (62.2%), greater than on the right with 222 (37.5%) cases. One (0.2%) patient with a rare median fissure was identified.

In the division into complete or incomplete, taking into account that the trans-foramen clefts are always classified as complete, the post-foramen cleft with the highest prevalence was incomplete in 257 patients (23.8%), whereas in the pre-foramen the cleft type more appeared in 139 patients (12.9%) (Table 2).

Table 2. Frequency of complete or incomplete forms compared between pre-, post- and trans-foramen clefts, in number (N) and percentage.

Fissure type	Complete		Incomplete		Total	
	N	%	N	%	N	%
Post-foramen	55	5.1	257	23.8	312	28.9
Pre-foramen	139	12.9	188	17.4	327	30.3
Trans-foramen	439	40.7	0	0	439	40.7
Total	633	58.7	445	41.3	1078	100

The most prevalent cleft in the general sample was unilateral trans-foramen with 307 individuals (28.5%) followed by incomplete post-foramen with 257 (23.8%). Rare fissures ($n = 2$) included alveolar fissures ($n = 1$) and Epignathus (median, $n = 1$) (Table 3).

Table 3. Frequency in number (N) and percentage (%) of the types of clefts.

Fissure type	N	%
Unilateral trans-foramen	307	28.5
Incomplete post-foramen	257	23.8
Incomplete unilateral pre-foramen	160	14.8
Bilateral trans-foramen	131	12.2
Complete unilateral pre-foramen	123	11.4
Complete post-foramen	55	5.1
Incomplete bilateral pre-foramen	27	2.5
Complete bilateral pre-foramen	16	1.5
Rare	2	0.2
Total	1078	100.00

When considering the anatomical structures that may be involved in the different types of fissures, the following aspects were established: to be characterized as a complete fissure, and it must be present in all the anatomical structures that may be involved; that is, the pre-foramen is complete when it affects the lip, dental arch up to the incisor foramen (12.9% of patients in the sample) and the post-foramen fissure affects from the incisor foramen to the end of the soft palate, in addition to the uvula (5.1%).

DISCUSSION

As for the malformation location in incomplete type fissures, the same structures are used, appearing partially or in isolation, as can be seen in Table 3. Among the affected anatomical references, one can perceive a predominance of exclusively lip involvement in 167 patients with an incomplete pre-foramen cleft. In 24 patients, only a red line was observed in the nasal filter together with the lip, called scar cleft. This scar can occur combined with cleft lip (2 cases in the sample) and other types of cleft, such as incomplete post-foramen (1 patient). When post-foramen was considered, there was a prevalence of isolated involvement of the soft palate (129 cases) followed by the hard palate partially associated with the soft palate (89 cases). Submucosal clefts appeared in 12 patients with post-foramen cleft and two patients with trans-foramen cleft without association with other soft tissues, and in 1 patient with an associated cleft lip. In the trans-foramen clefts, fissures were observed that affected all structures, from the lip to the soft palate's limits, having been observed in 368 patients (Table 4).

Table 4. Frequency in number regarding the location of the cleft related to the anatomical structures involved (in cases of incomplete clefts)

Specified location	Post	Pre	Trans	Total
Lip to soft palate			368	368
Isolated lip		167	25	192
Lip to incisive foramen		139		139
Isolated soft palate	129		4	133
Partial hard palate + soft palate	89		8	97
Incisive foramen to the soft palate	55			55
Partial soft palate	22		1	23
Muscular plane (submucosa)	12		2	14
Scar cleft		16	8	24
Lip + dental arch		2	10	12
Isolated uvula	3		2	5
Hard palate + soft palate	2		2	4
Scar fissure + lip + dental arch		1	1	2
Lip + partial hard palate + soft palate			3	3
Epignathus			1	1
Scar cleft + lip		2		2
Lip + partial hard palate + soft palate			1	1
Lip + soft palate			2	2
Lip + muscle plane			1	1
Total	312	327	439	1078

In our study, a higher general occurrence of cleft lip and/or palate was found in males (54.3%), following the literature that points to be a more common problem in this sex^{2,4}. However, still concerning sex, the female was, in absolute numbers, the most affected when it comes to post-foramen clefts. The literature shows that females are the most affected when it comes to cleft palate^{4,5}. The female predominance in isolated cleft palates is attributed to the earlier horizontalization of the palate in male embryos, which occurs after testicular differentiation. This would leave female embryos exposed to environmental factors for longer⁶.

Regarding the type of cleft, we found trans-foramen cleft (n = 439, 40.7%) and approximate values between post-foramen (n = 312, 28.9%) and pre-foramen (n = 327), 30.3%). The higher prevalence of trans-foramen cleft is in agreement with another study that used a similar classification⁷.

Regarding the clefts being unilateral or bilateral, not taking into account post-foramen clefts and rare for not having these classifications, we observed in our results a unilateral predominance in both sexes (77.2%). Regarding the side, there was a higher prevalence on the left side (62.3%). This finding also follows the literature that shows that unilateral cases are more common than bilateral and unilateral cases on the left^{4,5}.

The detailed description of all the structures affected by the fissure, besides the traditional classification itself, brings, in our view, some significant benefits. Details about the involvement of the lip, hard and/or soft palate, dental arch, uvula, and nasal filter increase the amount of information about the case, being useful in the record, especially in the surgical planning the execution of the procedure itself.

Since the etiology of CLP has not yet been fully elucidated and may be influenced by factors such as genetics and environmental exposure, among others, data referring to a medical center located in a given geographic area may also be of specific interest. As far as our research went, this is one of the few publications referring to CLP in our country's southern region, an area of almost 30 million people, as projected by the Instituto Brasileiro de Geografia e Estatística in 2018⁸. Also, in this region-specific geographic area, this research presents the largest number of patients affected.

As the main limitation of this study, we point out that only patients followed up at the medical center were included; therefore, it is not a broader population study, being indeed susceptible to unreported clinical cases, in addition to not being able to present a general prevalence of cases in the geographical area involved.

CONCLUSION

The most prevalent type of cleft was unilateral transforamen. In the female gender, the post-foramen cleft, and in the male, the transformation was the most frequent. Unilateral fissures were more common than bilateral fissures and, when unilateral, there was a predominance of lesions on the left. Regarding the anatomical details of the structures involved, the most common injuries were those that involved from the lip to the soft palate.

COLLABORATIONS

- JFP** Analysis and/or data interpretation, Conception and design study, Conceptualization, Final manuscript approval, Formal Analysis, Methodology, Project Administration, Supervision, Validation, Visualization, Writing - Original Draft Preparation, Writing - Review & Editing
- BSK** Conception and design study, Conceptualization, Data Curation, Final manuscript approval, Formal Analysis, Project Administration, Writing - Original Draft Preparation, Writing - Review & Editing
- EF** Analysis and/or data interpretation, Conception and design study, Data Curation, Final manuscript approval, Formal Analysis, Investigation, Writing - Original Draft Preparation, Writing - Review & Editing

RCT Analysis and/or data interpretation, Conception and design study, Final manuscript approval, Project Administration, Supervision, Writing - Original Draft Preparation, Writing - Review & Editing

RSF Analysis and/or data interpretation, Conception and design study, Final manuscript approval, Methodology, Project Administration, Supervision, Validation, Writing - Original Draft Preparation, Writing - Review & Editing

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