

Epidemiological profile of labiopalatine clefts in children cared for at a reference center in Paraná

Perfil epidemiológico das fissuras labiopalatais de crianças atendidas em um centro de referência paranaense

Perfil epidemiológico de las hendiduras labiopalatinas en niños tratados en un centro de referencia en Paraná

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Abstract: **Objective:** to analyze the epidemiological profile of labiopalatine clefts in children cared for at a care and research center on craniofacial anomalies. **Method:** this is an epidemiological, documentary and cross-sectional study that assessed 116 children, aged from 0 to 12 incomplete years, with labial and/or palatine cleft cared for at a reference center in western Paraná from February 2013 to February 2017 **Results:** there was a predominance of male patients (n=67; 57.8%), living in Cascavel (n=58; 50%) and over five years old at the time of the first consultation (n=41; 35.3%). The most frequent clefts were typified as: complete extension (n=56; 50.9%), labiopalatine (n=50; 43.1%) and left-sided unilateral (n=35; 44.3%). **Conclusions:** the acquired data are in accordance with the findings of the pertinent literature and show the need for measures that enable the early access of cleft children to this specialized service.

Descriptors: Child health; Cleft lip; Cleft palate; Epidemiology; Health profile

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Resumo: **Objetivo:** analisar o perfil epidemiológico das fissuras labiopalatais de crianças atendidas em um centro de atenção e pesquisa em anomalias craniofaciais. **Método:** estudo epidemiológico, documental, transversal, que avaliou 116 crianças, com idade de 0 a 12 anos incompletos, com fissura labial e/ou palatina atendidas em um centro de referência do oeste do Paraná de fevereiro de 2013 a fevereiro de 2017 **Resultados:** houve predominância de pacientes do sexo masculino (n=67; 57,8%), residentes em Cascavel (n=58; 50%) e com mais que 5 anos no momento da primeira consulta (n=41; 35,3%). As fissuras mais frequentes eram de extensão completa (n=56; 50,9%), do tipo labiopalatina (n=50; 43,1%) e unilaterais à esquerda (n=35; 44,3%). **Conclusões:** os dados obtidos estão de acordo com os achados da literatura e evidenciam a necessidade de medidas que viabilizem o acesso precoce das crianças fissuradas ao referido serviço especializado.

Descriptores: Saúde da criança; Fenda labial; Fissura palatina; Epidemiologia; Perfil de saúde.

Resumen: **Objetivo:** analizar el perfil epidemiológico de las hendiduras labiopalatinas en niños tratados en un centro de atención e investigación sobre anomalías craneofaciales. **Método:** estudio epidemiológico, documental, transversal que evaluó a 116 niños, de 0 a 12 años de edad, con labio leporino y/o palatino tratados en un centro de referencia en el oeste de Paraná desde febrero de 2013 hasta febrero de 2017 **Resultados:** hubo un predominio de pacientes masculinos (n=67; 57.8%), residentes en Cascavel (n=58; 50%) y mayores de 5 años en el momento de la primera consulta (n=41; 35.3%). Las fisuras más frecuentes fueron de extensión completa (n=56; 50,9%), del tipo labiopalatino (n=50; 43,1%) y unilaterales a la izquierda (n=35; 44,3%). **Conclusiones:** los datos obtenidos están en línea con los hallazgos de la literatura y muestran la necesidad de medidas que permitan el acceso temprano de los niños con hendiduras a ese servicio especializado.

Descriptores: Salud del niño; Labio leporino; Fisura del paladar; Epidemiología; Perfil de salud.

Introduction

Labial and/or palatine clefts are changes resulting from failures in the fusion of the nasal and maxillary formation processes, and they are considered as the most frequent facial congenital anomalies.¹ Such changes have wide variation, where labial, palatine, labiopalatine, unilateral or bilateral clefts are the most common, and can be found in separately or associated with other changes, as part of some syndrome.² Globally, the incidence of labiopalatine cleft is 0.5 to 2 cases for each 1,000 live births; and, in Brazil, the incidence is 1.0 case for each 650 live births.³

Patients with labiopalatine clefts, as well as their family members deal with several functional, psychological and social obstacles in the face of the diagnosis of malformation. The treatment of these patients should be held in a specialized interdisciplinary scope, aiming at esthetic, functional (suction, swallowing, chewing, breathing, speech, hearing) and psychosocial rehabilitation of the individual.³ In this context, the interaction of the multidisciplinary health team with the family and the patient is an indispensable action for the successful rehabilitation.⁴

By knowing the epidemiological profile of the patients cared for, we can enhance the actions of the health teams that assist them, as it enables us to reassess and guide the behaviors to be followed, allowing for a better structuring of the service. Accordingly, the research question was: what is the epidemiological profile of labiopalatine clefts in children cared for at a reference center in western Paraná? Thus, the present study was intended to analyze the epidemiological profile of labiopalatine clefts in children cared for at a care and research center on craniofacial anomalies.

Method

This is an epidemiological, documentary and retrospective study. The research was developed at the Care and Research Center on Craniofacial Anomalies (CEAPAC, as per its Portuguese acronym), which is attached to the University Hospital of Western Paraná (HUOP, as per its Portuguese acronym). The aforementioned service, opened in early 2013, offers specialized treatment for patients with labiopalatine clefts, mainly from the Western, Southwestern and

Northwestern Regions of Paraná (PR), as well as those originated from the States of Santa Catarina (SC) and Mato Grosso do Sul (MS), besides Paraguay (border region).

Until now, the State of Paraná has three reference centers for the treatment of patients with clefts, namely: the Comprehensive Care Center for Labiopalatine Cleft Patients (CAIF, as per its Portuguese acronym), located in the city of Curitiba; the Support and Rehabilitation Center for Labiopalatine Cleft Patients (CEFIL, as per its Portuguese acronym), located in the city of Londrina; and CEAPAC (previously mentioned), located in Cascavel. From February 2013 to February 2017, a total of 819 patients were registered at the aforementioned service, with 11,132 outpatient visits and 31 surgeries. Of this total, 4,778 assistance services and 19 surgical procedures were performed in children up to 12 years of age. This study was attended by 116 children who met the following selection criteria: having labial and/or palatine cleft, having from 0 to 12 incomplete years of age (11 years, 11 months and 29 days) at the time of the first consultation at the aforementioned service and having been cared for from February 2013 to February 2017.

The study variables were: gender; age at the first assistance service; origin; cleft specificities (type of cleft: labial [pre-foramen], labiopalatine [transforamen or pre/ post-foramen] and palatine [submucous or post-foramen]; extension of cleft: complete, incomplete and mixed; and laterality of cleft: right, left and bilateral). The cleft specificities were classified according to the authors based on the classification of Spina.²

Data were collected from the physical and electronic medical records (Tasy®) of the aforementioned service, entered into a Microsoft Office Excel® 2013, version 2.0, spreadsheet and exported to software R, version 3.6.0, for the accomplishment of statistical analyses. In order to verify the statistical association among the study

variables, we performed the Chi-square test and Fisher's exact test, considering a significance level of 5% ($\alpha=0.05$).

The research was approved by the Standing Research Ethics Committee with Human Beings of the State University of Maringá, opinion nº: 2.794.738/2018, CAAE: 85143717.0.0000.0104, approved on 08/01/2018.

Results

Of the 116 assessed patients (100%), 57.8% (n=67) were male; 50% (n=58) lived in Cascavel and 35.3% (n=41) were over five years old at the time of the first consultation at CEAPAC. The clefts most frequently identified in this study were: complete extension (n=56; 50.9%), left-sided unilateral (n=35; 44.3%) and labiopalatine type (n=50; 43.1) (Table 1).

Table 1 – Study variables of the children with labiopalatine clefts cared for at CEAPAC/HUOP from February 2013 to February 2017. Cascavel, 2018.

Variables	N	%
Gender*		
Male	67	57,8
Female	49	42,2
Origin*		
Cascavel	58	50,0
Other cities	58	50,0
Age at 1 st assistance service (months)*		
0 - 11	39	33,6
12 - 23	07	6,1
24 - 59	29	25,0
60 - 144	41	35,3
Type of Cleft*		
Labiopalatine	50	43,1
Labial	30	25,9
Palatine	36	31,0
Extension of Cleft**		
Complete	56	50,9
Incomplete	50	45,5
Mixed	04	3,6
Laterality of Cleft***		
Left	35	44,3
Right	25	31,6
Bilateral	19	24,1

*Size of sample: n=116

**Size of sample: n=110

***Size of sample: n=79

Statistical analyses showed that there was no association between the gender and origin of children and the cleft specificities (Table 2).

Table 2 – Demographic variables and cleft specificities of the children with labiopalatine clefts cared for at CEAPAC/HUOP from February 2013 to February 2017. Cascavel, 2018.

Variables	Diagnosis of Cleft																							
	Type*						Extension**						Laterality***											
	Labiopalatine		Palatine		Labial		Total	p§	Complete		Incomplete		Mixed		Total	p§	Left		Right		Bilateral	Total	p§	
	n	%	n	%	n	%			n	%	n	%	n	%			n	%	n	%	n	%		
Gender																								
Male	31	46,3	17	25,4	19	28,4	67	0,3029	33	49,3	31	46,3	3	4,5	67	0,7982	22	44,9	13	26,5	14	28,6	49	0,3373
Female	19	38,8	19	38,8	11	22,4	49		23	53,5	19	44,2	1	2,3	43		13	43,3	12	40,0	5	16,7	30	
Origin																								
Cascavel	20	34,4	19	32,8	19	32,8	58		23	42,6	29	53,7	2	3,7	54		18	46,2	15	38,5	6	15,4	39	
Other cities								0,1198								0,2198								0,1580
	30	51,7	17	29,3	11	19,0	58		33	58,9	21	37,5	2	3,6	56		17	42,5	10	25,0	13	32,5	40	

§Chi-square test

*Size of sample: n=116

**Size of sample: n=110

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This study also did not identify an association between the demographic variables (gender and origin) and cleft specificities (type, extension and laterality) and the child's age at the first assistance service performed at CEAPAC (Table 3).

Table 3 – Demographic variables and cleft specificities of the children with labiopalatine clefts according to age at the first assistance service performed at CEAPAC/HUOP, from February 2013 to February 2017. Cascavel, 2018.

Variables	Age at First Consultation (months)								Total	p§
	0 - 11		12 - 23		24 - 59		60 - 144			
	n	%	n	%	n	%	n	%		
Gender*										
Male	25	37,3	3	4,5	15	22,4	24	35,8	67	0,6850
Female	14	28,6	4	8,2	13	26,5	18	36,7	49	
Origin*										
Cascavel	13	22,4	4	6,9	18	31,0	23	39,7	58	0,0794
Other cities	26	44,8	3	5,2	11	19,0	18	31,0	58	
Type of Cleft*										
Labiopalatine	18	36,0	3	6,0	12	24,0	17	34,0	50	
Labial	7	23,3	2	6,7	7	23,3	14	46,7	30	0,8490
Palatine	14	38,9	2	5,6	9	25,0	11	30,5	36	
Extension of Cleft**										
Complete	20	35,7	3	5,4	14	25,0	19	33,9	56	
Incomplete	16	32,0	3	6,0	12	24,0	19	38,0	50	0,6433
Mixed	2	50,0	1	25,0	1	25,0	0	0,0	4	
Laterality of Cleft***										
Left	8	22,9	4	11,4	8	22,9	15	42,9	35	
Right	8	32,0	1	4,0	7	28,0	9	36,0	25	0,4347
Bilateral	9	47,4	0	0,0	4	21,1	6	31,6	19	

§Chi-square test

*Size of sample: n=116

** Size of sample: n=110

*** Size of sample: n =79

Discussion

In the assessed study sample (n=116; 100%), there was a predominance of male patients (n=67; 57.8%), living in Cascavel (n=58; 50%) and aged over five years at the time of the first consultation at CEAPAC (n=41; 35.3%) (Table 1). The greater number of cleft male patients is consistent with the findings in the pertinent literature.⁵⁻¹³ The fact that half of the assessed



patients (n=58; 50%) live in Cascavel does not mean that the number of patients with labiopalatine clefts is greater in this region. This finding may be explained by their easy access to CEAPAC and the possibility that other patients with labiopalatine clefts born in the countryside of Paraná have been referred to other reference services located in the State (CAIF and CEFIL).

The compliance with the current public health care policies¹⁴⁻¹⁵ that determine the establishment and accreditation of new specialized care centers for patients with craniofacial anomalies should favor the broad access of this population to prevention, treatment and rehabilitation services. The late arrival of children for treatment at CEAPAC (aged over five years) may be related to the short time of operation of this unit. As previously mentioned, CEAPAC was opened in February 2013, while the vast majority of patients cared for had already started their treatment at other specialized health centers.

As evidenced by the pertinent literature, the present study identified a predominance of labiopalatine (n=50; 43.1%),^{6,8-12,16-20} complete extension (n=56; 50.9%)^{6,10,19} and left-sided unilateral (n=35; 44, 3%) clefts.^{6,8,10,12,17,21} By knowing the profile of the predominant clefts in the surveyed service, we can contribute to the enhancement of the multidisciplinary therapeutic plan, since the more complex the injury, the greater the functional impairment of the assisted patient.¹⁰

There is still no consensus in the literature on the cause of the prevalence of cleft on the left side. An attempt of explanation would be the greater blood supply in the right hemiface during the embryonic period due to the blood pressure exerted by the right internal carotid.¹⁸ Another hypothesis would be the proximity of the affected region to the heart during the embryonic period, which could increase the risk for the formation of clefts due to the beat of the aforementioned organ.¹²

This study did not identify an association between the gender and origin of children and the cleft specificities. Although not statistically significant, male individuals showed the

highest proportions of labiopalatine ($n=31$; 46.3%),^{6,8,9,10,20} complete extension ($n=33$; 49.3%)^{6,18} and left-sided laterality ($n=22$; 44.9%) clefts.^{6,20} Also without statistical significance, patients born in Cascavel-PR showed higher proportions of incomplete extension ($n=29$; 53.7%), left-sided laterality ($n=18$; 46.2%) and labiopalatine type ($n=20$; 34.4%) clefts (Table 2).

Although there was no statistical association between the studied variables (Table 3), it is possible to make some considerations in this regard. As for the origin, we can observe that half ($n= 58$; 50%) of the children with labiopalatine clefts cared for at CEAPAC came from Cascavel-PR. This fact may be explained due to the exposure of pregnant women to polluting agents, common in large urban centers, which may contribute to the birth of children with clefts.²¹ The other half of the study sample ($n=58$; 50%) came from other smaller cities, with an essentially agricultural economy. In the same way the pollution acts, agrochemicals can favor the occurrence of congenital malformations in rural areas.²²

The data of this study show that the cities neighboring Cascavel ($n=26$; 44.8%) tend to refer children to CEAPAC earlier (in the first year of life) when compared to children originated from Cascavel ($n=13$; 22, 4%) (Table 3). This finding may be explained by the fact that small cities do not have assistance services that support children with special needs in the first months of life, thereby referring them to large centers earlier.

Although contradictory, the fact that most children born in Cascavel ($n=23$; 39.7%) take longer to start treatment at CEAPAC (after five years of life) may be explained by the variety of health care options available in the city, even if not specialized. Another hypothesis would be that, as it is a new health service (with less than four years of operation at the time of the collection of research data), most patients cared for at CEAPAC had already started treatment at other reference centers.

Although without statistical association, the patients with labiopalatine ($n=18$; 36.0%) and palatine (14; 38.9%) clefts, classified as mixed (pre and post-foramen) ($n=2$; 50.0%) and

bilateral (n=9; 47.4%) tend to reach the specialized service still in the first year of life. A justification for this would be the complexity of the aforementioned changes that may pose a risk to the maintenance of the child's life due to the difficulty in the feeding process.²³ Following this same reasoning, it is coherent to consider that, due to the less severe craniofacial change, children with labial type cleft (n=14; 46.7%) tend to reach the reference centers later (after five years of life).

The recommended age to start the treatment of the cleft patient is between one month and two years of age, due to the ideal surgical time for the primary lip and palate surgeries.^{10,24} The early search for treatment favors the achievement of more satisfactory esthetic and functional results because of the proper management of the therapeutic plan.²⁵ Accordingly, it is recommended a wide dissemination of specialized services in order to provide physical, emotional and social support both to the individual with craniofacial anomalies and to his/her family.¹³

Conclusions

The study identified a predominance of labiopalatine clefts in male patients (n=67; 57.8%), living in Cascavel (n=58; 50%), aged over five years at the time of the first consultation (n=41; 35.3%). The most frequent clefts were: complete extension (n=56; 50.9%), labiopalatine type (n=50; 43.1%) and left-sided unilateral (n=35; 44.3%).

The fact of having knowledge about the epidemiological profile of the assisted patients with labiopalatine clefts allows the planning of specific actions aimed at meeting the specificities of this population. The late access of children with labiopalatine cleft in the researched service reinforces the need for measures that enable their early admission to

specialized centers, since the early and suitable accomplishment of treatment provides more satisfactory esthetic, functional and psychosocial results.

We should highlight that the small number of patients that comprised the study sample could have been considered as a limiting factor of the research; however, the sample size corresponded to the totality of children cared for in the period of data collection. New research involving patients belonging to other age groups may assist in the guidance of the treatment of individuals with these craniofacial changes.

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