

# Analysis of a Facet of Lip and Palate Malformations in Brazil: A Decade-Long Ecological Study

# Análise de uma faceta das malformações labiopalatais no Brasil: Estudo ecológico de uma década

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# Abstract

Introduction Cleft lip and palate are malformations resulting from the nonunion or incomplete junction of facial development processes during embryonic life. Their categorization follows their embryological basis and relationships with the incisive foramen into three types: preforamen, postforamen, and transforamen. Surgical plastic interventions correct these congenital craniofacial deformities. Cheiloplasty is the cleft lip correction procedure and palatoplasty is the cleft palate correction surgery. As this is a severe public health problem, it is imperative to analyze the hospitalization stay periods and death rates resulting from this malformation in Brazil. **Materials and Methods** The present is an ecological study, that is, a retrospective, descriptive, and quantitative analysis of accessible public data from the Brazilian Unified Health System's (Sistema Único de Saúde, SUS, in Portuguese) Computer Sciences Department (Departamento de Informática do SUS, DATASUS, in Portuguese). The data source was the SUS Hospital Information System (Sistema de Informações Hospitalares do SUS, SIH/SUS) and the Live Birth Information System (Sistema de Informações Sobre Nascidos Vivos, SINASC, in Portuguese) on cleft lip and palate from 2014 to 2024. Data collection occurred in May 2024.

- cleft palate
  health
  Results In the last decade, Brazil had 73,829 hospitalizations and 69 deaths of patients with cleft lip and palate.
- medicine
  Surgery, plastic
  Conclusion This data supports the need for public policies on maternal and child health due to the proportion of this malformation in Brazil.

# Resumo

**Keywords** 

► cleft lip

Introdução As fissuras labiopalatais são malformações originárias da não junção ou junção incompleta dos processos de desenvolvimentos faciais durante a vida embrionária. São categorizadas por sua base embriológica e suas relações com o forame incisivo em três tipos: pré-forame, pós-forame e transforame. Essas deformidades craniofaciais congênitas são corrigidas por cirurgia plástica. Dessa forma, o procedimento de correção da fenda labial é denominado de queiloplastia, enquanto a cirurgia para a fenda palatina é chamada palatoplastia. Como esse é um grave problema de

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This is an open access article published by Thieme under the terms of the Creative Commons Attribution 4.0 International License, permitting copying and reproduction so long as the original work is given appropriate credit (https://creativecommons.org/licenses/by/4.0/) Thieme Revinter Publicações Ltda., Rua Rego Freitas, 175, loja 1, República, São Paulo, SP, CEP 01220-010, Brazil saúde pública, torna-se imperioso analisar a dimensão de internações e óbitos dessa malformação no Brasil.

**Materiais e Métodos** Este estudo representa uma pesquisa ecológica, caracterizada por uma abordagem retrospectiva, descritiva e quantitativa, que analisa dados públicos acessíveis do Departamento de Informática do Sistema Único de Saúde (DATASUS). A fonte utilizada é proveniente do Sistema de Informações Hospitalares do SUS (SIH/SUS) e o Sistema de Informações Sobre Nascidos Vivos (SINASC) sobre a fenda labial e palatina de 2014 a 2024. A coleta de dados foi realizada em maio de 2024. **Resultados** Durante a última década, o Brasil testemunhou um total de 73.829

Palavras-chave ► cirurgia plástica

- fenda labial
- fissura palatina
- medicina
- ► saúde

**Conclusão** A partir disso, são necessárias políticas públicas relacionadas a saúde materno-infantil, considerando a proporção dessa malformação no Brasil.

internações e 69 óbitos de pacientes com fenda labial e palatina.

## Introduction

Cleft lip and palate are anomalies with multifactorial etiology, involving genetic, environmental, or both factors, such as chromosomal alterations and maternal behavior in the prenatal period.<sup>1</sup> They arise due to failure in the fusion of facial processes during embryonic development. Lip and palate formation occurs between the fourth and twelfth weeks of gestation.<sup>2</sup> This fusion is crucial for the normal development of the face. In this critical period, small openings, known as foramen, allow communication between different developing facial structures. However, problems occurring in this process, such as tissue fusion interruptions, may result in malformations, such as cleft lip and palate.<sup>3</sup>

The landmark of the incisive foramen marks the distinction between the front and back clefts. Palate malformations are a result of the palatine shelves' lack of fusion due to their small size, their inability to elevate, inhibition of the fusion process, or the tongue's failure to settle between the shelves resulting from micrognathia. Meanwhile, cleft lips are defects caused by a partial or complete lack of fusion of the maxillary prominence with the medial nasal prominence on one or both sides.<sup>3</sup>

Although the classification of cleft lip and palate is diverse, the one proposed by Vitor Spina and under use in the plastic surgery service of Hospital das Clínicas da Faculdade de Medicina da Universidade de São Paulo (HCFMUSP), has a wide acceptance as it has an embryological basis to describe the deformities and their relationship with the incisive foramen. Malformations are categorized into three types: preforamen, postforamen and transforamen.<sup>4</sup> The literature reports the transforamen cleft as the most prevalent. It is worth mentioning a study in a pediatric hospital in northeastern Brazil, in which unilateral transforamen (24.7%) and complete postforamen clefts (23.2%).<sup>5</sup>

The diagnostic confirmation of these malformations can occur during prenatal care, at an ultrasound at the fourteenth week of gestation. Cheiloplasty consists of reconstructive surgery of the cleft lip and can occur when the patient is between 3 and 6-months-old. Palatoplasty reconstructs the cleft palate, occurring between 9 and 12-months-old.<sup>6</sup>

Surgical procedures for correcting cleft lip aim to restore the function and appearance of the lips and nose.<sup>7</sup> Two-flap palatoplasty is an advanced surgical technique for correcting complex cleft palates.<sup>8</sup> In addition to this technique, several other approaches can correct cleft palates. Furlow palatoplasty, a double reverse Z-plasty,<sup>2</sup> uses Z-flaps to lengthen the palate and reposition the levator veli palatini muscle and is effective in reducing velopharyngeal insufficiency.<sup>9</sup> Von Langenbeck palatoplasty, a traditional and simpler technique, involves elevating lateral mucoperiosteal flaps to enable central closure without tension. Additionally, Veau-Wardill-Kilner palatoplasty, or VY pushback, is a modification from the Von Langenbeck technique to lengthen the soft palate through lateral incisions, allowing palatal flap advancement.<sup>10</sup>

The rehabilitation process begins in childhood and extends into adulthood, requiring multidisciplinary treatment. The benefits of primary surgeries in childhood are evident, but they can result in a rigid and fibrous lip girdle in the maxillary region, hindering facial bone growth and maxillary development, and leading to teeth and facial deformities.<sup>11</sup>

Risk factors for cleft lip and palate include low birth weight, often associated with the choice of scheduled cesarean delivery. This can result in babies being born before full maturation, not reaching the final weight gain phase. Additionally, genetic factors, maternal smoking, and alcohol consumption during pregnancy significantly increase the risk of developing these conditions, according to the literature.<sup>12</sup>

# Objective

This article aims to analyze the number of hospitalizations and deaths, as well as calculate the mortality rate caused by cleft lip and palate in all Brazilian regions over a decade. The goal of this analysis is to provide a more in-depth understanding of this perinatal occurrence around the country. However, it does not represent its totality due to limitations in the data from the Brazilian Unified Health System's (Sistema Único de Saúde, SUS, in Portuguese ) Hospital Information System (Sistema de Informações Hospitalares do SUS, SIH/SUS).

# **Materials and Methods**

This study is ecological, that is, a retrospective, descriptive, and quantitative analysis of public data accessible through the TABNET application of the SUS's Computer Sciences Department (Departamento de Informática do SUS, DATA-SUS, in Portuguese ). The data source was the SIH/SUS and the Live Birth Information System (Sistema de Informações Sobre Nascidos Vivos, SINASC, in Portuguese). Data collection occurred in May 2024 and covered all reported cases of cleft lip and palate in Brazilian residents from 2014 to 2024. Since this study used secondary data, there was no requirement to submit it to the Ethics in Research Committee.

We organized the selected variables in tables using Microsoft Excel and presented them as absolute and relative numbers. The variables included the number of hospitalizations and deaths per region (North, Northeast, Southeast, South, and Midest). Additionally, the text presents the difference in hospitalization rates between genders, race/ethnicity, and the cost of hospital services and professionals for cleft lip and palate treatment. It is worth highlighting the sum of all live births from 2014 to 2024 is available on the Brazilian Ministry of Health's Integrated Health Surveillance Platform (Plataforma Integrada de Vigilância em Saúde, IVIS, in Portuguese), as well as and the percentage of hospitalized patients due to cleft lip.

Moreover, to support the data from SIH/SUS, we conducted searches on the PubMed and SciELO platforms using the descriptors *Cleft Lip* and *Surgery*, *Plastic* connected by the Boolean operator AND. SciELO retrieved three publications, and PubMed found 1,499. To filter the articles relevant to this research, the selection criterion was full articles published in the last 5 years. Exclusion criteria were theses, dissertations, incomplete articles, or irrelevant studies. It is also worth mentioning that we used plastic surgery books and reliable public-domain data. This strategy allowed for a more robust and contextualized analysis, enriching the understanding of the collected data and placing them within the broader context of existing research and scientific publications on cleft lip and palate in Brazil.

# Results

**- Table 1** shows the occurrence of 73,869 hospitalizations of patients with cleft lip and palate over the past decade in Brazil. The regions had different incidence rates, with the Southeast leading with 36,311 hospitalizations, followed by the Northeast, with 15,136. The North recorded 6,488 hospitalizations, while the South had 12,048. Finally, the Midwest presented 3,888 hospitalizations. These data from SIH/SUS of the Ministry of Health provided valuable insight

**Table 1** Hospitalization notifications due to cleft lip and palateby region from 2014 to 2024

Region	Hospitalizations
Total	73,869
North	6,488
Northeas	15,136
Southeast	36,311
South	12,046
Midwest	3,888

into the geographic distribution of hospitalizations for cleft lip and palate from February 2014 to 2024.

**- Table 2** shows that Brazil unfortunately recorded 69 deaths due to cleft lip and palate. These data from SIH/SUS of the Ministry of Health revealed an unequal distribution of deaths per region. The Southeast region led, with 21 deaths, followed by the Northeast region, with 16 deaths. The North recorded 14 deaths, while the South and Midwest regions had 10 and 8 deaths, respectively. This information provides significant insight into the magnitude and geographic distribution of deaths resulting from cleft lip and palate over a decade. The deaths may have occurred due to difficulties in the feeding process.

Thus, the total mortality rate in Brazil due to cleft lip and palate was 0.09 according to the SIH/SUS data from the Ministry of Health. These figures revealed a significant range in mortality rates by region. The Southeast recorded the lowest rate, with 0.06, followed by the South, with 0.07. On the other hand, the North and Midwest had higher rates, with 0.21 and 0.20, respectively. The Northeast presented a rate similar to the national average: 0.10.

According to data from the Ministry of Health, obtained through SUS/SIH, the total number of male hospitalizations due to cleft lip and palate in the last 10 years is 41,450, and 33,052 for female patients. In a regional analysis, the North recorded 3,778 male and 2,974 female hospitalizations. The Northeast had 8,347 male and 6,906 female hospitalizations. The Southeast presented the highest number in both categories, with 20,375 male and 16,133 female hospitalizations. The Southern region recorded 6,745 male and 5,320 female

**Table 2** Death notifications due to cleft lip and palate by regionfrom 2014 to 2024

Total69North14	Region	Deaths
North 14	Total	69
	North	14
Northeast 16	Northeast	16
Southeast 21	Southeast	21
South 10	South	10
Midwest 8	Midwest	8

hospitalizations, while the Midwest had 2,205 male and 1,719 female.

The distribution of hospitalizations due to cleft palate in Brazil, according to SUS/SIH data, revealed that 41.7% of admitted patients are white (31,037), followed by 32.9% mixed-race (24,568). Hospitalizations of black subjects represented 2.5% (1,883), while Asian and indigenous groups corresponded to 0.4% (305) and 0.3% (238), respectively. Additionally, 22.1% of hospitalizations (16,471) did not have information on color or ethnicity, indicating the need to improve data collection to understand better the prevalence of this malformation among the different Brazilian populations.

In the last 10 years, the value of hospital services for cleft lip and palate treatment in Brazil totaled R\$ 65,064,108.45. Most of this amount corresponded to the Southeast region, with a total of R\$ 31,957,409.38, followed by the Northeast, with R\$ 13,339,479.14. The South recorded expenditures of R \$ 11,304,139.59, while the North had R\$ 5,344,112.93 in expenses. Finally, the Midwest had the lowest amount, with R\$ 3,118,967.41 allocated to these services. These numbers reflect both the population distribution and the availability and demand for specialized treatment in each region.

In the last decade (2014–2024) the value of professional services for cleft lip and palate treatment in Brazil totaled R\$ 38,303,633.64 according to data from the SIH/SUS. The Southeast led the spending, with R\$ 18,871,975.98, followed by the Northeast, with R\$ 7,537,761.55. The Southern region recorded expenses of R\$ 7,365,067.38, while the North had R \$ 2,840,895.59. The Midwest had the lowest value, with R\$ 1,687,933.14 allocated to professional services for cleft lip and palate treatment. These values reflect the demand and availability of specialized professionals in each region throughout the period.

The IVIS platform revealed a total of 28,857,686 live births from 2014 to 2024. Thus, the number of hospitalizations due to lip and palate malformations is equivalent to 0.25% of live births.

#### Discussion

There are some significant limitations in SIH data-related research. Due to its nature, data collection occurs from hospitalized patients alone. As such, there is an inherent selection bias as data does not represent nonhospitalized subjects or those seeking treatment in other medical facility types.

Data quality can vary as it depends on the accuracy of the hospital records, potentially resulting in coding errors or missing information, compromising result validity. Moreover, geographic coverage of the data can undergo limitations by not including all regions or health facility types, affecting sample representativeness. However, SIH data remains a valuable source of information for public health and epidemiology research, as long as we recognize these limitations and adequately address them during result analysis and interpretation.<sup>13</sup> Analyzing the regions with the lowest report number of cleft lip and palate hospitalizations, their lower economic activity may explain the greater probability of underreported cases.<sup>14</sup>

Breastfed children with lip and palate malformations are subject to difficulties in the feeding process, which can lead to death. In cleft lip, this difficulty occurs due to the inadequate sealing of the baby's mouth around the areola. However, in cleft palate, the feeding difficulty results from loss of sucking strength.<sup>15</sup>

The number of cases of cleft lip and palate in Brazil, according to epidemiological data from SUS between 2014 and 2024, was higher in males. Research corroborates this trend, as a study from 2019 to 2023 demonstrated a significant predominance of hospitalizations among male patients, with 18,576 cases (55.27%), compared with 15,028 cases (44.72%) among females.<sup>16</sup> It is worth mentioning there is a previous study of the epidemiological profile of cleft lip and palate patients treated at a reference hospital in the countryside of Alagoas, in the Northeastern region, in which 67.7% of the 32 patients were male.<sup>17</sup>

Regarding the distribution by color/ethnicity, the present study demonstrated a higher prevalence in the White population. According to the literature, this prevalence is related to the higher frequency of the *IRF6* gene variant in subjects of European descent associated with these malformations.<sup>18</sup> In Brazil, they occur predominantly in White patients. However, other studies indicated a high incidence of anomalies in non-White children due to social inequality, as low-income strata have less access to healthcare services.<sup>19</sup> These results may be related to racial miscegenation in genotype definition.<sup>20</sup>

# Conclusion

The analysis of data on hospitalizations and deaths due to cleft lip and palate in Brazil over the last decade revealed the magnitude and geographic distribution of this condition. It is worth noting the high costs of hospital and professional services for cleft lip and palate correction and treatment in infants. In Brazil, most epidemiological studies on cleft lip and palate are specific, focusing on certain municipalities, states, regions, or healthcare services. There are few studies like this one, with population data at the national level. Thus, identifying regions with greater difficulties in managing and treating patients with cleft lip and palate can help develop more effective strategies to reduce mortality and improve the quality of life of the affected subjects. Therefore, it is crucial to continue monitoring these statistics and investing in resources and training for healthcare professionals, ensuring more equitable and efficient care throughout the country.

#### Authors' contributions

RSPdA: data analysis and/or interpretation, statistical analysis, final manuscript approval, funding acquisition, data collection, conceptualization, study conception and design, resource management, project management, investigation, methodology, performance of surgeries and/or experiments, writing – original draft, writing – review & editing, software, supervision, validation, and visualization.

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## **Clinical Trials?**

None.

#### **Conflict of Interests**

The author has no conflict of interests to declare.

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