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#### **ORIGINAL ARTICLE**

# TRENDS AND CLINICAL PROFILE OF LEPROSY CASES IN CHILDREN AND ADOLESCENTS UNDER 15 YEARS OLD IN INLAND BAHIA, BRAZIL

Alessandra Barboza Resende Martinez<sup>1</sup>, Júlio Martinez Santos<sup>2</sup>, Humberto Baptista Costa<sup>3</sup>, Maria Augusta Vasconcelos Palácio<sup>4</sup>, Maria Luisa de Carvalho Correia<sup>5</sup>, Iukary Takenami<sup>6</sup>

#### Highlights

- (1) From 2008 to 2021, the detection of childhood leprosy dropped from 20.1 to 3.8 cases/100,000.
  - (2) 31% of diagnosed children had prior contact with individuals with leprosy.
  - (3) Physical disability occurred in 12% of cases, including claw hand deformity.

#### **ABSTRACT**

Leprosy remains a significant public health challenge in Brazil, and its transmission among children and adolescents under 15 years old is predominantly associated with household contacts. Monitoring the leprosy cases within this specific age group serves as a crucial indicator for disease surveillance and control efforts. Thus, the aim of this study was to describe the trend and clinical profile of leprosy in children and adolescents under 15 years old in inland Bahia, Brazil. This retrospective cross-sectional follow-up study was performed in a specialized reference center in the Paulo Afonso municipality, Bahia. The data were obtained from the records of leprosy patients under 15 years, treated and followed up by the service between 2008 and 2022. The mean new case detection rate of leprosy in children and adolescents at 13.6 (95% CI: 7.9-19.4) cases per 100,000. The highest frequency was in the 10- to 14-year-old age group, comprising 62.1% of cases. Contact screening identified 31.0% of cases, and 12.3% had some disability at diagnosis. Paucibacillary cases (77.6%) were more prevalent than multibacillary, with tuberculoid clinical form (56.9%). The cure rate was 96.4%. The study highlights the need to intensify epidemiological surveillance efforts, including actively searching for cases, particularly among individuals aged 10 to 14 years, who represent the highest proportion of leprosy cases. Targeted interventions and education campaigns tailored to this age group are imperative to enhance awareness, early detection, and access to care, ultimately mitigating the burden of leprosy and preventing long-term sequelae among children and adolescents.

**Keywords:** adolescent health; child; epidemiology; health profile; leprosy.

#### TENDÊNCIA E PERFIL CLÍNICO DOS CASOS DE HANSENÍASE EM CRIANÇAS E ADOLESCENTES MENORES DE 15 ANOS NO INTERIOR DA BAHIA, BRASIL

#### **RESUMO**

A hanseníase ainda representa um desafio significativo para a saúde pública no Brasil, e sua transmissão entre crianças e adolescentes menores de 15 anos está predominantemente associada a contatos domiciliares. O monitoramento dos casos de hanseníase nesse grupo etário específico desempenha um papel crucial na vigilância da doença e nos esforços de controle. O objetivo deste estudo, portanto, foi descrever a tendência e o perfil clínico da hanseníase em crianças e adolescentes menores de 15 anos no interior da Bahia, Brasil. Trata-se de um estudo de seguimento transversal, retrospectivo, realizado em um centro de referência especializado no município de Paulo Afonso, Bahia. Os dados foram obtidos mediante consulta aos prontuários de pacientes com hanseníase menores de 15 anos, tratados e acompanhados pelo serviço entre 2008 e 2022. A taxa média de detecção de casos novos de hanseníase foi de 13,6 (IC 95%: 7,9-19,4) casos por 100.000 crianças e adolescentes. A maior frequência ocorreu na faixa etária de 10 a 14 anos, representando 62,1% dos casos. A triagem de contatos identificou 31,0% dos casos e 12,3% apresentaram algum grau de incapacidade física no momento do diagnóstico. Casos paucibacilares (77,6%) foram mais prevalentes que os multibacilares, com a forma clínica tuberculoide (56,9%). A taxa de cura foi de 96,4%. O estudo destaca a necessidade de intensificar os esforços de vigilância epidemiológica, incluindo a busca ativa de casos, especialmente entre indivíduos de 10 a 14 anos, que representam a maior proporção de casos de hanseníase. Intervenções direcionadas e campanhas educativas adaptadas a esse grupo etário são imperativas para aumentar a conscientização, a detecção precoce e o acesso aos cuidados, mitigando, assim, o ônus da hanseníase e prevenindo sequelas a longo prazo entre as crianças.

Palavras-chave: saúde do adolescente; criança; epidemiologia; perfil de saúde; hanseníase.

- <sup>1</sup> Universidade Federal do Vale do São Francisco (Univasf). Paulo Afonso/BA, Brasil. https://orcid.org/0000-0002-0053-434X
- <sup>2</sup> Universidade Federal do Vale do São Francisco (Univasf). Paulo Afonso/BA, Brasil. https://orcid.org/0000-0003-3486-8548
- <sup>3</sup> Universidade Federal do Vale do São Francisco (Univasf). Paulo Afonso/BA, Brasil. https://orcid.org/0009-0001-4122-5279
- <sup>4</sup> Universidade Federal do Vale do São Francisco (Univasf). Paulo Afonso/BA, Brasil. https://orcid.org/0000-0003-3373-0264
- <sup>5</sup> Serviço de Dermatologia e Pneumologia Sanitária (Sederpas/SMS). Paulo Afonso/BA, Brasil. https://orcid.org/0000-0002-2780-125X
- <sup>6</sup> Universidade Federal do Vale do São Francisco (Univasf). Paulo Afonso/BA, Brasil. https://orcid.org/0000-0001-5660-7766



### INTRODUCTION

Leprosy is a chronic infectious disease caused by a slow-growing bacterium named *Mycobacterium leprae*, which manifests itself through dermatological and neural lesions<sup>1</sup>. More recently, *Mycobacterium lepromatosis* was identified as a new species and the second causal agent of the disease<sup>2</sup>. According to the World Health Organization (WHO), in 2021, 140,594 new cases of leprosy were reported worldwide, with 9,052 of those cases occurring in children and adolescents. Brazil alone reported 18,318 new cases, of which 4.1% (761) were in individuals under 15 years of age<sup>3</sup>. In comparison, in 2020, there were approximately 128,405 new cases reported, with 8,629 occurring in children and adolescents<sup>4</sup>.

The spatial distribution of leprosy in Brazil is extremely heterogeneous, with emphasis on pockets of poverty located in the Midwest, North, and Northeast regions, respectively. The state of Bahia, located in the Northeast region, identified 1,537 new cases of leprosy in 2021, with an estimated detection rate of 10.3 per 100,000 inhabitants. In the population under 15 years of age, 46 new cases were reported, and the detection rate was 1.47 per 100,000 inhabitants, which is considered a pattern of medium endemicity for this age group<sup>5</sup>.

The detection of leprosy in children and adolescents under 15 years old is an important indicator of active and recent transmission in the community. The occurrence of leprosy cases among children and adolescents suggests the existence of bacilliferous adults who have not been identified by the health system and, consequently, have not received treatment<sup>5,6</sup>. This epidemiological scenario encourages discussions about possible failures associated with diagnosis, active patient searching, screening of household contacts, and health education practices<sup>5</sup>.

Furthermore, the diagnosis of leprosy in children and adolescents tends to be more difficult, mainly due to the difficulty in applying and correctly interpreting sensitivity tests, increasing the possibilities of complications and deformities. A study by Souza, Luna, and Magalhães<sup>7</sup> showed that there is a trend of late diagnosis of leprosy in Bahia compared to other states in Brazil. This delay between the appearance of signs and symptoms and the diagnostic confirmation contributes to the emergence of disabilities and physical deformities. According to Moraes et al.<sup>8</sup>, 20% of children had some degree of physical disability at diagnosis, which has a negative impact on their social life and can have significant effects on their personality in adulthood. Thus, as there is no vaccine capable of preventing the development of the disease, early diagnosis is still considered the most important tool in eliminating and coping with the disease<sup>9</sup>.

Considering the particularities and implications of the disease in children and adolescents under 15 years old, it is necessary to develop studies that support improvements in public policies and epidemiological surveillance actions with an emphasis on the local profile of users of the unified health system. Additionally, studying the clinical features of leprosy in children and adolescents can help clinicians improve early diagnosis and develop better treatment protocols, ultimately reducing the risk of physical deformities and disabilities. Thus, given the pivotal role of epidemiology in supporting authorities in guiding public health actions, and in alignment with the objectives outlined in the Global Strategy for Leprosy 2021-2030, which aims to reduce the global and local burden of leprosy to zero<sup>5</sup>, the study aimed to describe the trend and clinical profile of leprosy in children and adolescents under 15 years old in inland Bahia, Brazil.



# **METHODS**

# Study design and setting

This is a retrospective cross-sectional follow-up study based on records of leprosy cases under 15 years old diagnosed or referred to the Dermatology and Sanitary Pneumology Service (SEDERPAS), in the municipality of Paulo Afonso, Bahia, between 2008 and 2022. The health unit serves as a referral center for the municipality of Paulo Afonso and neighboring regions, providing specialized services for comprehensive care in the management of leprosy and tuberculosis.

The municipality has a territorial area of 1,463 km<sup>2</sup> and it is located in the north of the state of Bahia, 480 km away from the state capital. The estimated population for 2021 was 119,213 inhabitants, of which 78.4% resided in the urban area, distributed across 64 neighborhoods, and 23.5% were children and adolescents under 15 years old<sup>10</sup>.

# Study population and ethical considerations

The diagnosis of leprosy cases was made according to the criteria established by the World Health Organization (WHO) and the Brazilian Ministry of Health<sup>5,11</sup>. This included an epidemiological analysis, dermatoneurological examination to identify lesions or areas of skin with altered sensitivity and/or peripheral nerve involvement (sensory, motor, and/or autonomic), and bacilloscopy of the intradermal smear, when available<sup>5,11</sup>. In compliance with the requirements of Resolution 466/2012 of the Brazilian National Health Council, this study was approved by the Research Ethics Committee of the Federal University of São Francisco Valley (Univasf), under opinion nº 3,830,208.

# Data collection

The data were collected by two observers and tabulated in an Excel® spreadsheet (Microsoft® Office 2003, Boston, MA, USA), validating them by double typing. The variables used in this study were: sex, age, race, year of diagnosis, clinical form, operational classification, detection mode, degree of disability, number of lesions, nerves involvement, bacilloscopy result, and treatment outcome. After data collection, re-entry cases or duplicate/illegible data were excluded from database.

The new case detection rate of leprosy was calculated by dividing the number of cases detected in a year by the total population in that year, and then multiplying the result by 100,000. The total population was consulted at the Brazilian Institute of Geography and Statistics (IBGE), based on the 2010 population census of the municipality and with intercensus projections for the age group from 5 to 14 years. Then, the detection rate among those under 15 years of age were classified as hyperendemic ( $\geq$  10.00); very high (9.99 to 5.00); high (4.99 to 2.50); medium (2.49 to 0.50); and low (< 0.50), according to the classification used by the Brazilian Ministry of Health<sup>12</sup>.

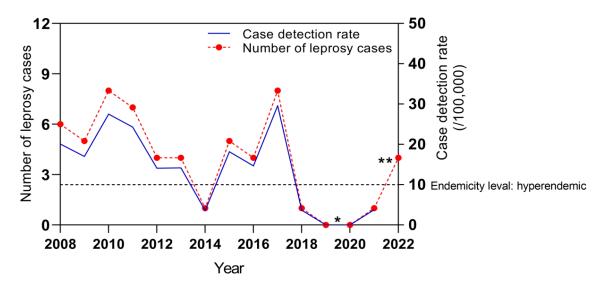
### Statistical analysis

The data were analyzed using the software GraphPad Prism v.8 (GraphPad Software, San Diego, CA, USA). The descriptive statistical measures used were absolute value (n), relative frequency (%), means and standard deviation (SD) or 95% confidence interval (95% CI). Descriptive analyzes of the data were performed by Chi-squared and Fisher's Exact test to evaluate the association between categorical variables. The Pearson's correlation test (r) was used to measure the correlation between continuous variables. Differences were considered statistically significant when the p-value less than 0.05.



# **RESULTS**

During the 15-year study period, a total of 58 new cases of leprosy were diagnosed in children and adolescents under 15 years old. The mean new case detection rate was calculated to be 13.6 (95% CI: 7.90-19.4) cases per 100,000. There was a decrease in the rate from 20.1 cases in 2008 to 3.8 cases per 100,000 children and adolescents in 2021. Notably, there were no registered cases of leprosy among individuals under 15 years of age in the years 2019 and 2020. The annual distribution of leprosy cases and detection rates is visualized in Figure 1.



<sup>\*</sup>No cases were registered in the years 2019 and 2020.

Figure 1 – Annual distribution of leprosy cases and detection rates among children and adolescents under 15 years old in Sederpas, Paulo Afonso, Bahia, from 2008 to 2022.

Source: Research data.

The mean age of study population was  $10 \pm 2.6$  years with 36 (62.1%) individuals aged between 10-14 years. Thirty-five (60.3%) girls were identified and 45 (78.9%) declared themselves as brown-skinned. Eighteen (31.0%) of them reported contact with diagnosed case of leprosy and therefore were detected by contact screening. The vast majority did not undergo/ignored bacilloscopy (29/58, 50.0%). However, among those who underwent, 89.7% (26/29) tested positive bacilloscopy. In addition, 45 (77.6%) cases had fewer than five lesions and six had some nervous impairment (Table 1). There was no observed correlation between the number of lesions and the age of the child (r = 0.037, p = 0.78). Similarly, no correlation was observed regarding the number of affected nerves (r = 0.080, p = 0.53).

The distribution of leprosy according to operational classification was as follows: 77.6% for paucibacillary cases, which corresponds to the predominant clinical forms; 20.7% for indeterminate cases; and 56.9% for tuberculoid cases (Table 1). The indeterminate lesions appeared as hypochromic spots with slight decrease in sensitivity (Figure 2A), while the lesions in cases with the tuberculoid form were characterized by well-defined annular erythematous plaque with elevated external borders with significant change in sensitivity (Figure 2B).

<sup>\*\*</sup>The detection rate for 2022 was not calculated due to the unavailability of population data from Brazilian Institute of Geography and Statistics (IBGE).



Seven (12.3%) individuals presented some degree of physical disability at the time of diagnosis, with four and three being diagnosed with grade 1 and grade 2, respectively. Among the observed deformities, the claw hand was particularly noteworthy, as shown in Figure 3.

Table 1 – Sociodemographic and clinical characteristics of leprosy cases in children and adolescents under 15 years old in Sederpas, Paulo Afonso, Bahia, from 2008 to 2022

Variables	N	%
Gender		
Male	23	39.7
Female	35	60.3
Age		
0-9 years	22	37.9
10-14 years	36	62.1
Race (self-declared) (N=57)		
White	9	15.8
Brown	45	78.9
Black	3	5.3
Detection mode		
Referral	20	34.5
Spontaneous demand	17	29.3
Community screening	2	3.4
Contact screening	18	31.0
Other modes	1	1.7
Operational classification		
Paucibacillary	45	77.6
, Multibacillary	13	22.4
Clinical form		
Indeterminate	12	20.7
Tuberculoid	33	56.9
Borderline	11	19.0
Lepromatous	2	3.4
Number of cutaneous lesions		
1-5 lesions	45	77.6
≥ 6 lesions	13	22.4
Bacilloscopy (N=29)		
Negative	3	10.3
Positive	26	89.7
Disability (N=57)		
Grade 0 disability	50	87.7
Grade 1 disability	4	7.0
Grade 2 disability	3	5.3
Peripheral nerves involvement		
No	52	89.7
Yes	6	10.3
Treatment completion (N=55)*		
No	2	3.6
Yes	53	96.4

 $<sup>\</sup>hbox{*Among the three cases that are currently unavailable, two are still undergoing treatment.}$ 

Source: Research data.





Figure 2 – Indeterminate leprosy: hypochromic spots with indefinite borders (A). Tuberculoid leprosy: well-defined annular erythematous plaque (B).

Source: Research data.



Figure 3 – Claw hand deformity observed in an 11 years-old child, showing significant bone resorption in the index finger.

Source: Research data.



# DISCUSSION

The early detection of leprosy in children is crucial for preventing the transmission of the disease and reducing morbidity and disabilities among this group. Although previous studies have already demonstrated a high frequency of leprosy cases<sup>13,14</sup>, this study provides novel insights by focusing specifically on children and adolescents under 15 years old in a municipality located in the interior of the state of Bahia. Our study unveiled important observations regarding the detection rate of leprosy in this population, highlighting the persistent presence of the disease in children and adolescents. This emphasizes the importance of not only continuing to search for existing cases but also actively identifying children and adolescents in contact with bacilliferous patients.

Despite a national decline in leprosy cases<sup>15</sup>, including among children and adolescents, the elimination of the disease in certain municipalities remains a pressing and challenging public health issue. Between 2008 and 2022, our findings revealed that the municipality exhibited detection rates above 10 cases per 100,000 inhabitants. This threshold indicates hyperendemic for leprosy among children and adolescents under 15 years old within<sup>12</sup>. Consequently, the Paulo Afonso municipality can be classified as a hyperendemic region, given the mean new case detection rate in this study of 13.6 cases per 100,000 children and adolescents. This suggests the persistence of *M. leprae* transmission and possible failures in detecting and treating bacilliferous adults both inside and outside households. In addition, the real potential for transmission from extra-household contacts remains unknown<sup>6</sup>.

However, the detection rate remained below 10 cases per 100,000 inhabitants at some points: 2014 and the period from 2018 to 2021. It is plausible to consider that the decline in the detection rate during those years could be attributed to underreporting and/or operational failures in contact surveillance, as supported by Ribeiro et al.<sup>16</sup> in the Diamantina microregion of Minas Gerais state. Consequently, it is crucial to acknowledge that the observed fluctuation in the detection rate may have been influenced more by operational factors rather than purely epidemiological factors.

The age group with the highest frequency was found to be between 10 to 14 years old, corroborating with other studies conducted in the states of Tocantins<sup>17</sup> and Pernambuco<sup>18</sup>, as well as the study by Sales et al.<sup>19</sup>, which evaluated the North and Northeast regions. Since the incubation period of the bacillus can range from two to seven years, it is often anticipated that the onset and detection of the disease would be more common in older children<sup>17</sup>. Additionally, the longer exposure time due to age may have also contributed to the increase in cases in this age group. Although not significant, an upward linear trend between the number of lesions and age can be observed. Moreover, delays and difficulties in diagnosis are also factors that may have contributed to the increase in the detection rate among children close to adolescence<sup>20</sup>.

Our study revealed a higher detection rate in girls (60.3%), in agreement with the national proportion of diagnosed cases according to the last Epidemiological Bulletin released in 2022, which reported 51.5% of cases were in girls<sup>15</sup>. Furthermore, this pattern of leprosy among women is also observed in the adult population<sup>14,21</sup>. The high prevalence of leprosy cases among children and adolescents of mixed race (78.9%) in all age groups in the region can be attributed to the extensive racial mixing within the population. Furthermore, an elevated occurrence of leprosy has previously been observed among adults of mixed race in the same municipality<sup>13,14</sup>. This demographic group often experiences unfavorable socioeconomic conditions, including limited access to healthcare services, education, and basic sanitation.

A total of 31.0% of the leprosy cases were diagnosed through contact examination of patients who had previously diagnosed with the disease. These findings highlight the effectiveness of active case finding in achieving higher detection rates<sup>22</sup>. However, delays in seeking care often result in a significant gap between the onset of symptoms and diagnosis, resulting in delayed treatment.



Household contacts represent the primary source of leprosy transmission, although transmission can occur beyond immediate family members, encompassing anyone in regular and close proximity to an untreated individual. The transmission of leprosy to children serves as an epidemiological indicator that the disease is still actively circulating within communities. The fact that children continue to contract leprosy underscores the need for enhanced and more efficient public health initiatives to effectively halt the spread of this disease.

Interestingly, 29.3% of new cases were detected through spontaneous demand by individuals, which may indicate a higher level of awareness about the disease among the population or could also reflect care-seeking based on parental concern. Alternatively, it could suggest a reliance on this pathway for diagnosis, resulting in detection only in more advanced stages of the disease<sup>23,24</sup>.

The paucibacillary form of leprosy, characterized by up to five skin lesions and no or minimal nerve involvement, was the most prevalent form found in the study. However, 22.4% of children and adolescents under 15 years old were classified as having the multibacillary form. This classification indicates that these children presented with more than five skin lesions, involvement of more than one nerve, or positive skin smears in any location, indicating a high bacillary load<sup>25</sup>. The detection of children and adolescents with the multibacillary form emphasizes the importance of maintaining vigilant and effective surveillance to identify and treat cases of the disease at an early stage. Early detection of these cases in children is particularly crucial, as the disease can progress rapidly, leading to irreversible physical disabilities.

The polychemotherapy regimen has demonstrated significant efficacy in reducing physical disabilities, deformities, and the disease transmission within the community, particularly when implemented early. However, the study revealed that 12.3% of children and adolescents presented with some form of disability at the time of leprosy diagnosis, potentially attributed to delayed detection, indicating deficiencies in the early case detection system<sup>5,20</sup>. A similar study conducted in Tocantins reported a disability prevalence of 9.5% in children and adolescents under 15 years old<sup>17</sup>.

The WHO aims to eliminate grade-two disabilities in children and adolescents under 15 years old, highlighting the need to comprehend the factors contributing to the persistence of disabilities or deformities at the time of diagnosis<sup>5</sup>. In terms of treatment outcomes, the observed cure rates were higher (96.4%) compared to another study conducted in the state of Mato Grosso between 2001 and 2013, where the cure rate was approximately 40.0%<sup>26</sup>. These findings underscore the effectiveness of health services in ensuring treatment adherence until full recovery. In the context of leprosy treatment, achieving a cure rate above 90.0% is regarded as exceptional and highly commendable.

Among the study's limitations, it is important to consider that the information used was obtained through a consultation of the Sederpas database, and possible errors resulting from typing and recording, as well as underreporting, may have occurred. Despite this, it is believed that because it is official data from the health unit, its results have achieved the proposed objectives.

# CONCLUSION

Leprosy remains a significant public health concern that requires coordinated efforts at all levels of government. This study's high detection rate indicates that bacillus transmission is still active in the region, emphasizing the urgent need for local authorities to implement targeted interventions to reduce the disease burden, particularly among priority areas and specific population groups such as individuals aged 10 to 14. While the cure rate is promising, the presence of multibacillary cases and physical disabilities highlights the challenges in controlling and eliminating leprosy. Therefore, additional research is essential to identify risk factors in this age group and develop effective preventive strategies that can have a more substantial impact on the local population.



Furthermore, the findings suggest the importance of expanding intradomiciliary contact examinations, which can facilitate early detection and prompt treatment to break the transmission chain of the bacillus. Enhancing surveillance efforts, implementing targeted interventions, and prioritizing early detection and treatment, it is possible to reduce the disease burden and prevent long-term complications associated with leprosy.

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#### **Author contributions**

**Alessandra Barboza Resende Martinez:** Conceptualization, Data acuration, Formal analysis, investigation, Methodology, Visualization, Writing – original draft.

Júlio Martinez Santos: Data acuration, investigation, Visualization, Writing – original draft.

**Humberto Baptista Costa:** Data acuration, investigation, Visualization, Writing – original draft.

Maria Augusta Vasconcelos Palácio: Formal analysis, Investigation, Methodology, Writing – review & editing.

**Maria Luisa de Carvalho Correia:** Formal analysis, Investigation, Methodology, Project administration, Supervision, Visualization, Writing – review & editing.

**lukary Takenami**: Conceptualization, Formal analysis, investigation, Methodology, Project administration, Supervision, Visualization, Writing – review & editing.

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### **Corresponding author**

Iukary Takenami
Universidade Federal do Vale do São Francisco (Univasf)
Av. da Amizade, 1900, CEP 48605-780, Paulo Afonso/BA, Brasil. iukary.takenami@univasf.edu.br



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