

Short Communication

Physical disabilities due to leprosy in Alagoas State, Northeast Brazil: a temporal and spatial modeling

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Abstract

Introduction: Monitoring disability trends is required to evaluate leprosy elimination. We assessed the trends in disability indicators and its association with space in Alagoas, Brazil. **Methods:** We conducted an ecological study in all leprosy cases from 2006 to 2016. Disability indicators were analyzed using the joinpoint regression model. **Results:** The proportion of new cases with grade 2 disability at diagnosis ranged from 4.7% to 11.9% (annual percent change, 1.4; $P=0.7$), while at discharge, it ranged from 0.0% to 12.3% (annual percent change, -21.8; $P=0.2$). **Conclusions:** Disability indicators had a stable trend over the study period.

Keywords: Brazil. Leprosy. Monitoring. Physical disability.

Leprosy is a chronic infectious disease caused by *Mycobacterium leprae* that affects the skin and peripheral nerves and, if untreated, can result in physical disabilities and/or deformities¹. An estimated two million people worldwide are currently living with disabilities or deformities due to leprosy². In Brazil, the proportion of new leprosy cases with grade 2 disability (G2D) in 2017 was 8.3%, and the rate was 9.39 per 1 million population³. However, disability is unevenly distributed across the country, with the highest proportion of cases reported in the north and northeast regions. In 2017, the detection coefficient of new leprosy cases in Alagoas was 9.06 cases per 100,000 population, and the G2D rate was 6.1 patients per 1 million population³.

Because of the high number of people with disability/deformity due to leprosy, the World Health Organization (WHO) has recommended the adoption and strengthening of strategies to reduce the disease burden and the prevalence of deformities⁴.

Therefore, monitoring the incidence and disability trends of leprosy is needed to evaluate the progress of these. Thus, this study assessed the trends of physical disability indicators in Alagoas State from 2006 to 2016.

Hence, we conducted an ecological study in all leprosy cases in Alagoas State from 2006 to 2016. Alagoas consists of ~ 3.3 million populations, and it is divided in 102 municipalities. The municipality was considered as the unit of analysis, and we examined the trend of the WHO leprosy indicators⁴.

Data were obtained from two Brazilian information systems. Information on the clinical and demographic characteristics of leprosy patients was obtained from the Notifiable Diseases Information System (SINAN-Sistema de Informações de Agravos de Notificação, in Portuguese)(<http://www2.datasus.gov.br/DATASUS/index.php?area=0203&id=31032752>), and the absolute annual population was obtained from the Brazilian Institute of Geography and Statistics (IBGE-Instituto Brasileiro de Geografia e Estatística, in Portuguese)⁵.

From the clinical and population data, we calculated the WHO leprosy indicators following the WHO⁴ and Brazilian Ministry of Health (MoH) guidelines⁶. The WHO leprosy indicators were the following: (a) rate of new cases with G2D at diagnosis per 1 million population, (b) proportion of new

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cases with G2D at diagnosis (high, $\geq 10\%$; medium, 5% to 9.9%; low, $< 5\%$), (c) proportion of new cases with G2D at discharge (high, $\geq 10\%$; medium, 5% to 9.9%; low, $< 5\%$), (d) proportion of new cases with physical disability assessed at diagnosis (good, $\geq 90\%$; regular, $\geq 75\%$ to 89.9%; poor, $< 75\%$), and (e) proportion of cases with physical disability assessed at discharge (good, $\geq 90\%$; regular, $\geq 75\%$ to 89.9%; poor, $< 75\%$).

The study population was described using frequencies and percentages. The trends of the WHO leprosy indicators were analyzed using the joinpoint regression model with the Monte Carlo permutation method⁷, which allowed us to identify the trends and change points for each WHO leprosy indicator and to determine the annual percentage change (APC). Additionally, each trend was categorized as stable, increasing, or decreasing. A significance level of 5% and a 95% confidence interval were used. The Joinpoint Regression Program 4.5.0.1 (National Cancer Institute, USA) was used in this analysis.

We examined the spatial dependence of each WHO leprosy indicator using the global Moran and the pseudo-significance tests. The Moran index ranges from -1 to +1, with negative values indicating a negative autocorrelation, positive values a positive autocorrelation, and values close to zero a randomization. When a spatial dependency was identified, we also used the local index of spatial association (LISA) method to detect regions with significant spatial correlations. According to the results of the LISA method, a Moran scattering diagram was generated, in which each municipality was positioned in a quadrant according to its value. Q1 (high/high) and Q2 (low/low) represent neighboring municipalities with similar values, indicating positive spatial association; Q3 (high/low) and Q4 (low/high) represent neighboring municipalities with distinct values, indicating negative spatial association. Based on the results obtained from the Moran and LISA scatter plot, thematic Moran maps were created. A *P* value less than 0.05 was considered statistically significant. Terra View 4.2.2 (Instituto Nacional de Pesquisas Espaciais) and QGIS 2.14.11 (Open Source Geospatial Foundation) were used in these analyses.

This study did not require approval from the ethics committee as it was using open public domain data without the identification of the individuals.

A total of 4,252 new leprosy cases were diagnosed in the general population in Alagoas from 2006 to 2016, and 1,284 (30.2%) individuals had disabilities at diagnosis, with 967 (22.7%) grade 1 disability (G1D) and 317 (7.5%) G2D. At discharge, 683 (18.3%) patients had disabilities, with 501 (13.4%) G1D and 182 (4.9%) G2D.

The rate of new leprosy cases with G2D ranged from 5.1 to 13.4 per 1 million population, with the highest and lowest rates being observed in 2008 and 2009, respectively. The average rate was 9.1 per 1 million population over the period (**Figure 1A**). Despite this variation, the joinpoint analyses identified a stable trend (APC, -3.1; *P*=0.4) (**Table 1**). The proportion of new cases with physical disability assessed at diagnosis remained over 83% (**Figure 1B**) and had no significant variations throughout the study period (APC, -0.3; *P*=0.3) (**Table 1**), while the proportion of new cases with G2D at diagnosis ranged from

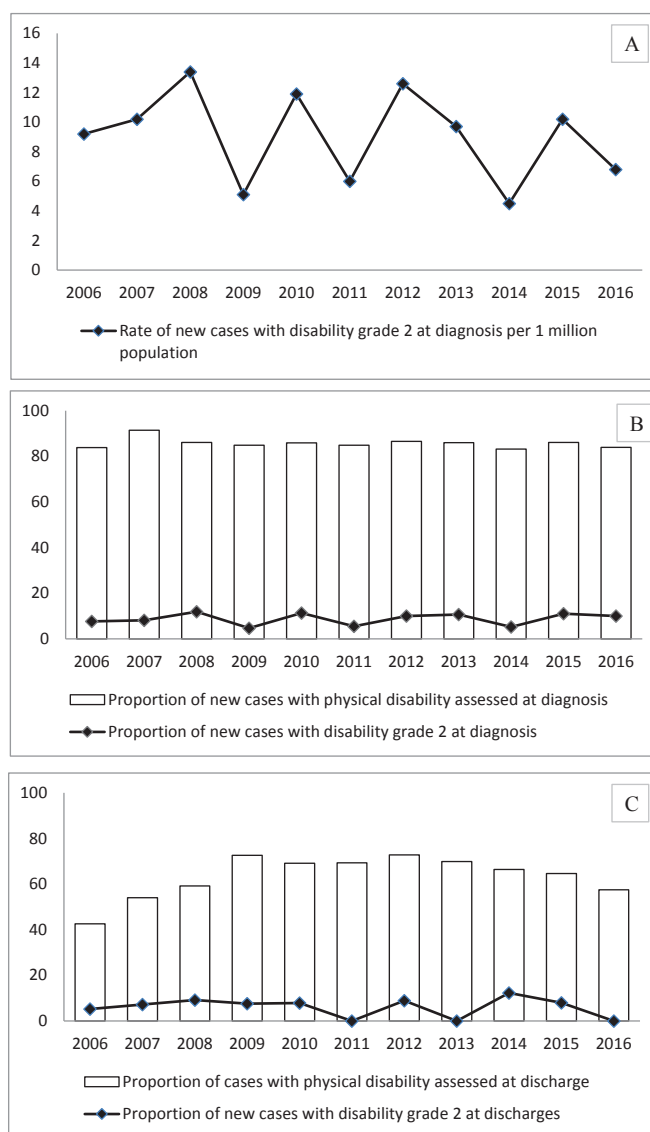


FIGURE 1: Epidemiological indicators related to leprosy, according to the year of notification in the state of Alagoas, Brasil, 2006-2016.

4.7% to 11.9% (APC, 1.4; *P*=0.7, considered a stable trend) (**Figure 1B** and **Table 1**). Conversely, the proportion of patients with physical disability assessed at discharge was less than 73% (**Figure 1C**), with a significant variation of 3.7% (*P*<0.001) (**Table 1**). The proportion of cases with a G2D at discharge ranged from 0.0% to 12.3% (APC, -21.8; *P*=0.2, considered a stable trend) (**Figure 1C**). Additionally, the joinpoint regression model identified an increasing trend in G2D between 2006 and 2009 (APC, 19.3; *P*<0.001), followed by a stable trend from 2009 (APC, -2.4; *P*=0.1).

Figure 2 shows the spatial distribution of the WHO leprosy indicators with respect to physical disability. Fifty (49.0%) of the 102 municipalities in Alagoas State did not register new cases of leprosy with G2D over the study period, although all municipalities reported leprosy cases over the study period. Of the 52 municipalities with reported cases of physical disability, 11 (10.8%) had rates between 1.0 and 4.9, 16 (15.7%) had rates

TABLE 1: Joinpoint regression analysis of the WHO leprosy indicators in Alagoas State, Northeast Brazil, from 2006 to 2016.

Indicator	Period	APC	95% CI	P value
Rate of new cases with disability grade 2 at diagnosis per 1 million population	2006–2016	-3.1	-10.7 to 5.2	0.4
Proportion of new cases with disability grade 2 at diagnosis	2006–2016	1.4	-6.0 to 9.5	0.7
Proportion of new cases with disability grade 2 at discharge	2006–2016	-21.8	-49.1 to 20.3	0.2
Proportion of new cases with physical disability assessed at diagnosis	2006–2016	-0.3	-0.8 to 0.3	0.3
	2006–2009	19.3	8.8 to 30.9	<0.001
Proportion of cases with physical disability assessed at discharge	2009–2016	-2.4	-4.8 to 0.0	0.1
	2006–2016	3.7	1.0 to 6.4	<0.001

APC: Annual Percent Change.

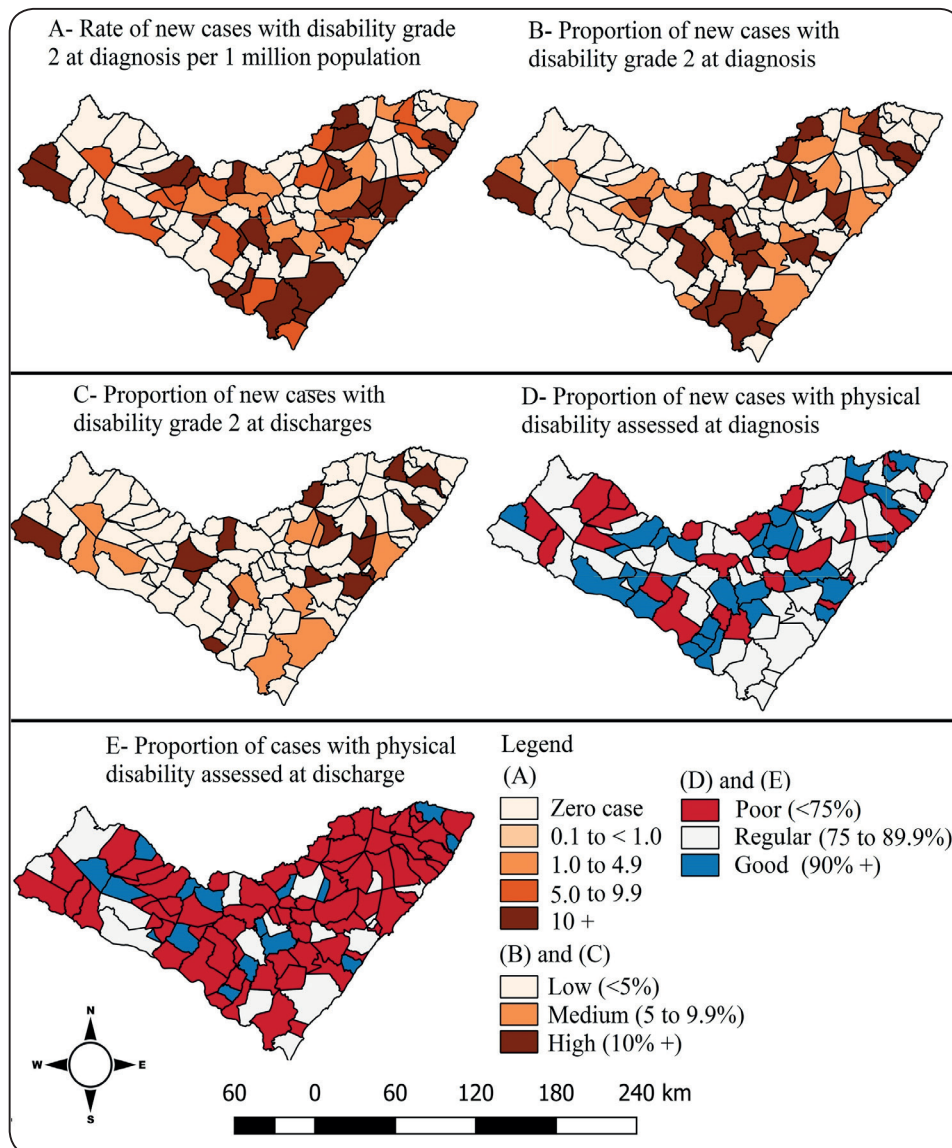


FIGURE 2: Maps of the epidemiological indicators of leprosy in Alagoas State, Northeast Brazil, from 2006 to 2016.

between 5.0 and 9.9, and 25 (24.5%) had rates greater than 10 cases per 1 million population. There was no evidence of spatial dependence of this indicator (I Moran = 0.02; $P = 0.2$).

Regarding the proportion of new cases with G2D at diagnosis, 58 (58.6%) municipalities had a low proportion (including the silent municipalities) according to the Brazilian MoH, 17 (16.6%) a moderate proportion, and 27 (24.8%) a high proportion (**Figure 2B**). At discharge, 75 (73.5%) municipalities had a low proportion of disabled individuals (less than 5%), 10 (9.8%) municipalities had a moderate proportion, and 17 (16.7%) municipalities had a high proportion. There was no spatial dependence at either of the two times (diagnosis/discharge) (I Moran = 0.07; $P = 0.1$ and I Moran, -0.007; $P = 0.3$, respectively) (**Figure 2C**).

At diagnosis, 37 (36.3%) municipalities evaluated the degree of physical disability in greater than 90% of patients, 41 (40.2%) municipalities in between 75% and 90% of patients; and 24 (23.5%) municipalities in less than 75% of cases, respectively (**Figure 2D**). Only 16 (15.7%) of the 102 municipalities assessed disability in greater than 90% of cases, 17 (16.7%) in between 75% and 90% of cases, and 69 (67.6%) in less than 75% of cases (**Figure 2E**). The proportion of new leprosy cases with disability assessed at diagnosis had a homogeneous spatial distribution (I Moran, -0.007; $P = 0.4$) and a heterogeneous distribution at discharge (I Moran, -0.145; $P = 0.006$).

The WHO leprosy indicators on physical disability are commonly used by control programs to monitor and evaluate the epidemiological scenario of leprosy, to reveal the changes in the transmission chain, and to make inferences with respect to the quality of the health care services^{4,8,9}. This study evaluated the trend of the WHO physical disability leprosy indicators in Alagoas using historical data from over a period of 11 years. Our findings revealed deficiencies in the evaluation of new cases of leprosy with disabilities at both the time of diagnosis and discharge. Moreover, despite the relatively low proportion of leprosy patients being evaluated for physical disability compared to the international recommended parameters⁴, the data revealed a high proportion of cases with G2D at both diagnosis and discharge⁶. These findings indicate the persistence of leprosy transmission and likely reflect operational control problems and poor-quality health care services available to eliminate the disease.

The average rate of leprosy cases with G2D was high (9.1 per 1 million population) in Alagoas State, similar to the rates found in other Brazilian regions³ and far from the target of 1 per 1 million population recommended by the WHO⁴. Additionally, trend analyses revealed a steady pattern in the timeline *continuum*, suggesting the persistence of disease transmission in Alagoas; hence, it will be difficult to achieve the WHO targets in the short- or medium-term, particularly if the same control actions continue to be performed. Therefore, we suggest that active case detection and actions to improve contact tracing should be strengthened.

The high proportion of new leprosy cases being diagnosed with G2D reinforces our concerns about leprosy in Alagoas, since these indicators also reflect operational problems and

likely reflect barriers in accessing healthcare services. As leprosy has a long incubation period, the presence of late presenting signs of disabilities at the time of the diagnosis indicates that the disease was already in its late stage. Earlier diagnosis is likely established in areas with better primary care services¹⁰⁻¹². Moreover, the high proportion of leprosy cases with G2D reinforces the evidence that the transmission chain is being maintained in the community, since, in general, leprosy patients with visible disabilities present advanced forms of the disease (e.g., multibacillary leprosy).

However, the proportion of disability due to leprosy in Alagoas may be even higher, given the low number of patients examined at both diagnosis and discharge, suggesting improper patient management, lower ability in assessing the presence of disability, inadequate monitoring of neural functions throughout the treatment, and lack of actions performed to prevent the occurrence of disability using the healthcare services^{11,12}. In this respect, the quality of health care services needs to be improved, with proper training for healthcare professionals so that they are able to establish an early diagnosis and adequately manage patients in order to avoid deformities, which result in social stigma, social isolation, and poor quality of life, due to leprosy^{2,13,14}.

Our findings also raise some concerns about the hidden prevalence of leprosy in Alagoas, which has been demonstrated in other studies¹⁵. In some regions with a leprosy prevalence of less than 1 case per 10,000 population, indicating that the disease has been eliminated according to the WHO parameters, the registration of individuals disabled by leprosy has remained high, suggesting that the disease has not actually been eliminated. This reinforces the need for improvements in control programs and healthcare services to search for cases of leprosy in the community. Additionally, development of more accurate tools in the early detection of cases of leprosy is necessary.

Our study has some limitations, which means its findings should be interpreted carefully. As the data were obtained from information systems, it represents information of patients who sought treatment, and consequently, there may be a number of cases in the community not recorded by the healthcare personnel. Additionally, there was a high proportion of leprosy patients with an unknown degree of physical disability, which may have had an effect on the WHO leprosy indicators used in this study. However, major efforts have been made to minimize any introduction of bias. Furthermore, our findings and their interpretation are supported by 11 years of information on leprosy reported in Alagoas.

In conclusion, our findings reveal that the indicators for disability had a stable trend throughout the period, with a relatively low number of leprosy patients being assessed for disabilities. Additionally, there was a high proportion of individuals with G2D at both diagnosis and discharge in Alagoas. Such findings indicate the persistence of leprosy transmission, late diagnosis, operational control problems, and the poor quality of healthcare services available to eliminate the disease. Additionally, the proportion of G2D cases raises concerns about the hidden prevalence of the disease and reinforces the need to strengthen the available control programs.

Conflict of interest

The authors have no conflict of interest to declare.

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