ORIGINAL ARTICLE / *ARTIGO ORIGINAL*

Low performance of operational indicators for leprosy control in the state of Bahia: spatiotemporal patterns, 2001–2014

Baixo desempenho de indicadores operacionais de controle da hanseníase no estado da Bahia: padrões espaçotemporais, 2001–2014

Eliana Amorim de Souza[I] (ID), Jorg Heukelbach[II,III] (ID), Maria Leide Wand-Del-Rey Oliveira[IV] (ID), Anderson Fuentes Ferreira[II] (ID), Sebastiao Alves de Sena Neto[V] (ID), Marcos Tulio Raposo[VI] (ID), Alberto Novaes Ramos Jr.[II] (ID)

ABSTRACT: *Objective:* To characterize spatiotemporal patterns of operational indicators for leprosy control in the state of Bahia from 2001 to 2014. *Methods:* This is a population-based ecological study, with spatial distribution and autocorrelation of operational indicators for leprosy control. *Results:* From 2001 to 2007, 42.7% (n=178) of the municipalities presented a cure rate lower than 75%, increasing to 61.4% (n =291) from 2009 to 2014. Between 2001 and 2007, 32.5% (n=54) of the municipalities reported more than 10% of the total number of relapses in the state, increasing to 36.9% (n=75) between 2008 and 2014. From 2001 to 2014, 38% (n=159) of the municipalities presented an assessment index of disability grading at the time of diagnosis within the regular performance parameter. Between 2009 and 2014, the number of municipalities with a high incidence of grade 2 disability (G2D) at the time of diagnosis increased, reaching 55.3% (n=230) of the municipalities. Most municipalities in the state of Bahia showed poor performance in the implementation of planned actions for leprosy control, with little change or relative worsening in the patterns of operational indicators throughout the historical series. *Conclusion:* The operational context in Bahia indicates significant institutional vulnerability, leading to the need for expansion and qualification of the surveillance and health care network in the different regions and conditions analyzed in the public health system (*Sistema Único de Saúde* – SUS).

Keywords: Leprosy. Epidemiology. Surveillance. Brazil.

[I]Multidisciplinary Health Institute, Universidade Federal da Bahia, Campus Anísio Teixeira – Vitória da Conquista (BA), Brazil.

[II]Department of Community Health, School of Medicine, Universidade Federal do Ceará – Fortaleza (CE), Brazil.

[III]College of Public Health, Medical and Veterinary Sciences, Division of Tropical Health and Medicine, James Cook University – Townsville, Queensland, Australia.

[IV]Universidade Federal do Rio de Janeiro, Hospital Universitário Clementino Fraga Filho – Rio de Janeiro (RJ), Brazil.

[V]Leprosy Control Program, State Secretariat of Health – Porto Velho (RO), Brazil.

[VI]Department of Health, Universidade Estadual do Sudoeste da Bahia – Jequié (BA), Brazil.

Corresponding author: Eliana Amorim de Souza. Instituto Multidisciplinar em Saúde, Universidade Federal da Bahia, Campus Anísio Teixeira. Rua Hormindo Barros, 58, Candeias, CEP: 45029-094, Vitória da Conquista, BA, Brazil. E-mail: eliana.amorim@ufba.br

RESUMO: *Objetivo:* Caracterizar padrões espaçotemporais de indicadores operacionais de controle da hanseníase no estado da Bahia no período de 2001 a 2014. *Metodologia:* Estudo ecológico, de base populacional, com distribuição e autocorrelação espacial de indicadores operacionais da hanseníase. *Resultados:* No período de 2001 a 2007, 42,7% (n = 178) dos municípios apresentaram percentual de cura inferior a 75%, ampliando para 61,4% (n = 291) de 2009 a 2014. De 2001 a 2007, 32,5% (n = 54) dos municípios notificaram mais de 10% do total de casos de recidiva do estado, com aumento para 36,9% (n = 75) dessa situação no período de 2008 a 2014. De 2001 a 2014, em 38% (n = 159) dos municípios, o indicador de avaliação do grau de incapacidade física (GIF) no momento do diagnóstico se encontrava conforme o parâmetro regular de desempenho. Já no período de 2009 a 2014, houve aumento de municípios com alta ocorrência de GIF2 no momento do diagnóstico, atingindo 55,3% (n = 230) dos municípios. A maioria dos municípios do estado da Bahia apresentou desempenho insatisfatório na execução das ações de controle previstas para a hanseníase, com pouca mudança ou relativa piora nos padrões de indicadores operacionais ao longo da série histórica. *Conclusão:* O contexto operacional do estado da Bahia sinaliza graves dimensões de vulnerabilidade institucional, o que implica necessariamente a ampliação e a qualificação da rede de vigilância e atenção à saúde nas diferentes regiões e contextos analisados do Sistema Único de Saúde (SUS).

Palavras-chave: Hanseníase. Epidemiologia. Vigilância. Brasil.

INTRODUCTION

Leprosy is a chronic condition with high potential to cause physical disabilities due to neural damage, in addition to esthetic damage and stigma. It is a persistent public health problem in Brazil, presenting a heterogeneous and focal distribution in areas with different levels of endemicity[1-3].

Leprosy affects mainly people in situations of significant vulnerability. Its neglected disease nature gives leprosy relative priority in the implementation of political agendas to tackle the condition in terms of health surveillance, control, care, and education[4-6]. Its control involves the development of intersectoral actions to fight poverty and social inequalities so as to reduce its incidence[7,8], in addition to structuring an integral and effective care network for people affected[9].

In Brazil, individuals, families, and communities affected or at risk of acquiring leprosy should find its main *locus* of care in primary health care (PHC)[4,9]. Since PHC is closer to people's everyday lives, including their family dynamics and household and social contact networks, it has a greater potential to identify and meet health needs, integrating the development of surveillance actions to interrupt transmission in the territories[9].

Due to its slow development and prolonged treatment, leprosy requires follow-up to promote adherence to the treatment prescribed, prevent and monitor adverse events and leprosy reactions, encourage self-care, prevent and rehabilitate physical disabilities, intensify the active search for new cases, and implement the surveillance

of new cases and their contacts, among other[1,4,10]. However, operational difficulties need to be overcome to ensure the performance of these actions in the different realities of the country[11].

Aiming to analyze and monitor the actions developed to control the disease, the Ministry of Health (MoH) recommends the use of operational indicators that allow evaluating the quality of the follow-up performed by health services, including the proportion of patients discharged as cured or who discontinued their treatment during the cohort years and the proportion of new cases of relapse. Relapse can indicate therapeutic failure, as well as reinfection and difficulty in differentiating leprosy reactions from relapses among health professionals[4].

Considering the natural history of leprosy, assessing the individual's neural function is essential, given the inherent risk of infection by *Mycobacterium leprae*. Thus, operationalizing disability grading becomes strategical[3,4]. Disability grading can vary between 0, 1, and 2, with grade 2 disability (G2D) corresponding to more severe cases. The proportion of patients who had their disability graded at the time of diagnosis and the number of them who presented G2D indirectly measure the coverage and quality of actions for the timely detection of cases and the monitoring of neural damage[4,11].

In this scenario, the present study aimed to characterize the spatiotemporal patterns of operational indicators for leprosy control in the state of Bahia from 2001 to 2014. It includes the 417 municipalities of Bahia and uses space as a category of analysis to provide significant information for surveillance in the territories[12].

METHODS

STUDY DESIGN AND DATA SOURCES

This is a population-based ecological study, with spatiotemporal analysis of operational indicators for leprosy control. We used secondary data from the Notifiable Diseases Information System (*Sistema de Informação de Agravos de Notificação* – SINAN) of the Brazilian MoH. The study included new leprosy cases notified from 2001 to 2014, excluding those that had "misdiagnosis" as the exit criterion. Georeferencing of spatial information and analysis was based on digital maps of the municipalities of Bahia provided by the National Institute for Space Research (*Instituto Nacional de Pesquisas Espaciais* – INPE).

This study was part of a doctoral dissertation in Public Health presented by Eliana Amorim de Souza for the Graduate Program in Public Health at Universidade Federal do Ceará (PPGSP-UFC) in 2017. The project was submitted to the Research Ethics Committee of Universidade Federal do Ceará (Certificate of Presentation for Ethical Consideration – *Certificado de Apresentação para Apreciação Ética* – CAAE: 19258214.2.0000.5054) and approved under report number 544,962.

STUDY SITE

We included all 417 municipalities in the state of Bahia. With an estimated population of 15 million inhabitants for 2015, Bahia is part of a group of states with the worst socio-demographic indicators in the country, particularly regarding the high income concentration[13,14]. Administratively, the state is organized in nine health districts[15].

STATISTICAL ANALYSIS

We calculated the following operational indicators, according to MoH guidelines[4]:
- Cure: proportion of leprosy cure among the new cases diagnosed in the cohort years;
- Discontinuation: proportion of leprosy patients who discontinued their treatment among the new cases diagnosed in the cohort years;
- Relapse: proportion of relapses among the cases notified during the year;
- Disability grading: proportion of new leprosy cases with disability graded at the time of diagnosis;
- G2D at diagnosis: proportion of leprosy cases with G2D at the time of diagnosis among the new cases detected and evaluated in the year under analysis[4].

We calculated the indicators for the proportion of cure and treatment discontinuation by considering the cohort from 2003 to 2014, as recommended by the MoH[4]. The remaining indicators were calculated from 2001 to 2014. Disability grading followed the guidelines established by the MoH, as well as parameters used to assess each indicator[4]. For cases of relapse, however, the parameters set for territorial analysis were: absence of cases of relapse, <5%, 5 to 10%, and >10%. We emphasize that, due to operational issues, we did not analyze the relapse research form, as recommended by the MoH. The variable under analysis corresponds to the field 'case type' in the form for notification of leprosy cases and includes all those registered as relapse.

Data were combined into municipality centroids to distribute the indicators spatially. We established the following intervals: 2001–2007, 2008–2014, and total period. The intervals specified for the indicators calculated in the cohort were: 2003–2008, 2009–2014, and total period. We chose these intervals because we did not identify relevant changes in smaller ones, resulting in analyses with greater consistency. We calculated the indicators by using the total number of cases for each period. The databases were consolidated in the TabWin® software, version 4.2, of the Technology Department of the public health system (*Departamento de Informática do Sistema Único de Saúde* – DATASUS-MS – http://datasus.saude.gov.br/apresentacao).

We adopted the local Moran method (local index of spatial association – LISA), which compares the indicators for each municipality with those of its neighbors, to identify possible areas and patterns of spatial autocorrelation. For the analysis of the proportion of cure

among new cases and disability graded at diagnosis, we calculated the proportion of new "non-cured" leprosy cases and new cases with "disability not graded at diagnosis." The analysis of these indicators was inverted to facilitate spatial visualization and interpretation of the patterns identified.

We employed the approach used by Moran Maps to construct the maps, considering municipalities with a statistically significant difference. General statistical analyses were conducted in the Stata software, version 11.2 (StataCorp LP, College Station, TX, USA). The software ArcGIS, version 9.3 (Environmental Systems Research Institute – ESRI, Redlands, CA, USA), and TerraView, version 4.1 (INPE, São José dos Campos, SP, Brazil), were used to process, analyze, and present cartographic data, as well as calculate indicators for spatial autocorrelation.

RESULTS

Out of a total of 31,688 new leprosy cases that comprised the cohort from 2003 to 2014, 85.2% were discharged as cured. From 2001 to 2007, 42.7% (n=178) of municipalities presented a cure rate lower than 75%. Between 2009 and 2014, this number increased, reaching 61.4% (n=291). All nine health districts in the state had municipalities with this operational pattern (Figure 1A)

Out of all cohort cases, 4,689 were not cured. While comparing the proportion of non-cured cases in each municipality with its neighbors, we found a spatial autocorrelation. From 2003–2008, the southwest district showed an important cluster that disappeared from 2009–2014. On the other hand, new clusters emerged in the east-central and south districts during this period. Between 2003 and 2014, the clusters in the southwest and east-central districts remained (Figure 1B).

In the cohort analyzed, 2,730 people had discontinued their treatment, with an annual average of 5.5%. A high percentage of municipalities did not register cases of treatment discontinuation (n= 278; 66.7%). Among those that did (n=139; 33.3%), most of them had less than 10 cases (n=79; 56.8%) reported. From 2009–2014, the number of municipalities in this scenario increased (n=154; 77.9%). The west, east-central, and southwest districts showed the highest number of municipalities with treatment discontinuation (Figure 2A). The spatial autocorrelation indicated the clustering of municipalities from 2003 to 2008, with high discontinuation rates in the southwest and east-central districts. Between 2009 and 2014, the concentration was more expressive in the southwest, south, and northeast districts of the state (Figure 2B).

Bahia presented a significant number of notifications of relapse, with an annual mean rate of 3.6% of cases in the period. From 2001–2007, a high number of municipalities reported cases of relapse (n=166; 40.0%). Among them, 32.5% (n=54) notified more than 10% of the total number of cases, located mainly in the north, west, and far-south districts. From 2008 to 2014, the number of municipalities with notification of relapse increased; 36.9%

of them (n=75) reported more than 10% of cases, particularly in the north-central district (Figure 3A). Between 2001 and 2007, few municipalities showed a spatial association for relapse in the south district of the state. In the following period, new clusters emerged in the southwest and east-central districts. The map for the entire period indicates the presence of small clusters in the south, southwest, and east-central districts (Figure 3B).

The mean percentage of new cases with disability graded at diagnosis was 86.2%. Throughout the period, 9.9% (n=3,967) of patients did not have their disability graded, and in 3.6% (n=1,427) of cases, this information was ignored, as the specific field in the SINAN notification form was not filled. From 2001–2007, 41% (n=171) of municipalities had 90% or more new cases with disability graded at the time of diagnosis. This percentage increased to 48% from 2008 to 2014. However, between 2001 and 2014, 38% (n=159) of municipalities had this indicator within the regular performance parameter (≥75 to
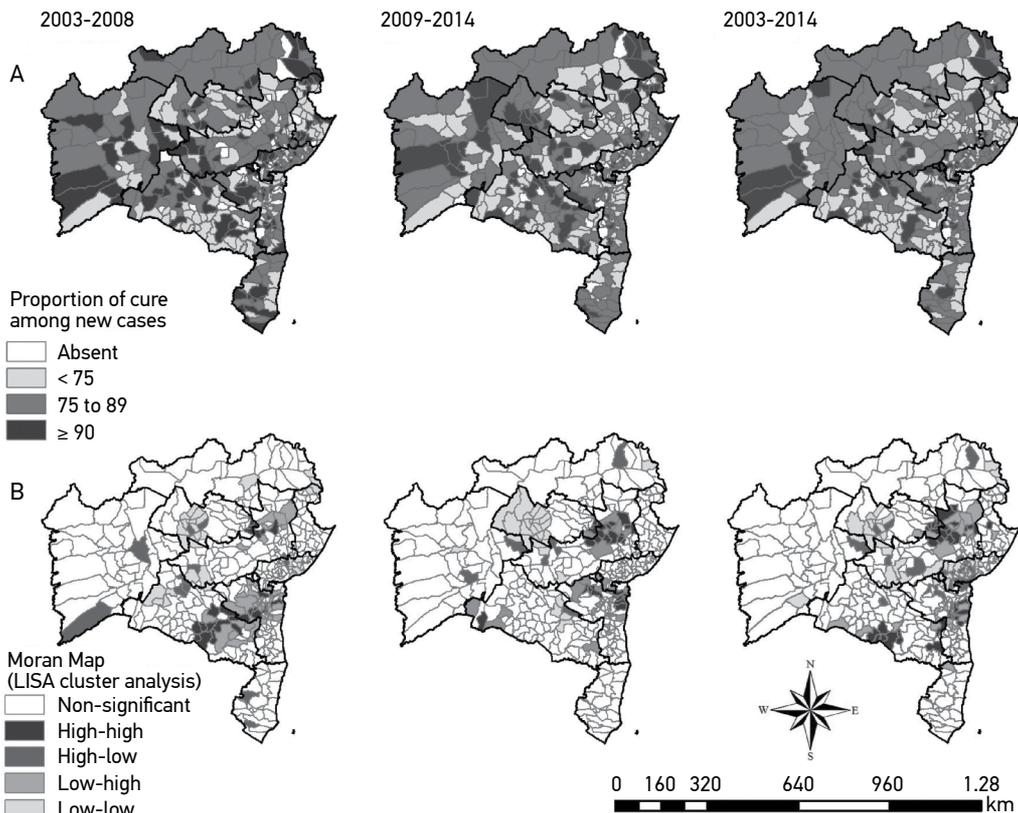

Figure 1. Spatial distribution of the proportion of leprosy cure among new cases diagnosed in the cohort years according to (A) municipality and spatial autocorrelation and (B) local Moran for the proportion of non-cured cases in the cohort. Bahia, 2003–2008, 2009–2014, and 2003–2014.

89.9%), and 15% (n=65), within the poor performance parameter (<75%). We found a greater concentration of these municipalities in the west, far-south, and east-central districts (Figure 4A).

The maps relating to the local Moran (Figure 4B) showed clusters of cases without disability graded at diagnosis, especially in the north and south districts of the state, all involving a small number of municipalities (2001–2007). The map for the entire period (2001–2014) confirmed the presence of clusters with poor performance in these same regions.

Among the cases that had their disability graded at the time of diagnosis, 64.4% (n=25,797) were classified as G0D, 17.3% (n=6,942) as G1D, and 4.8% (n=1,921) as G2D. The mean proportion of cases categorized as G2D at the time of diagnosis in the historical series was 5.7%. The low proportion of disability graded at the time of discharge from the multidrug therapy (MDT) prevented the construction of this indicator following the MoH guidelines[4].
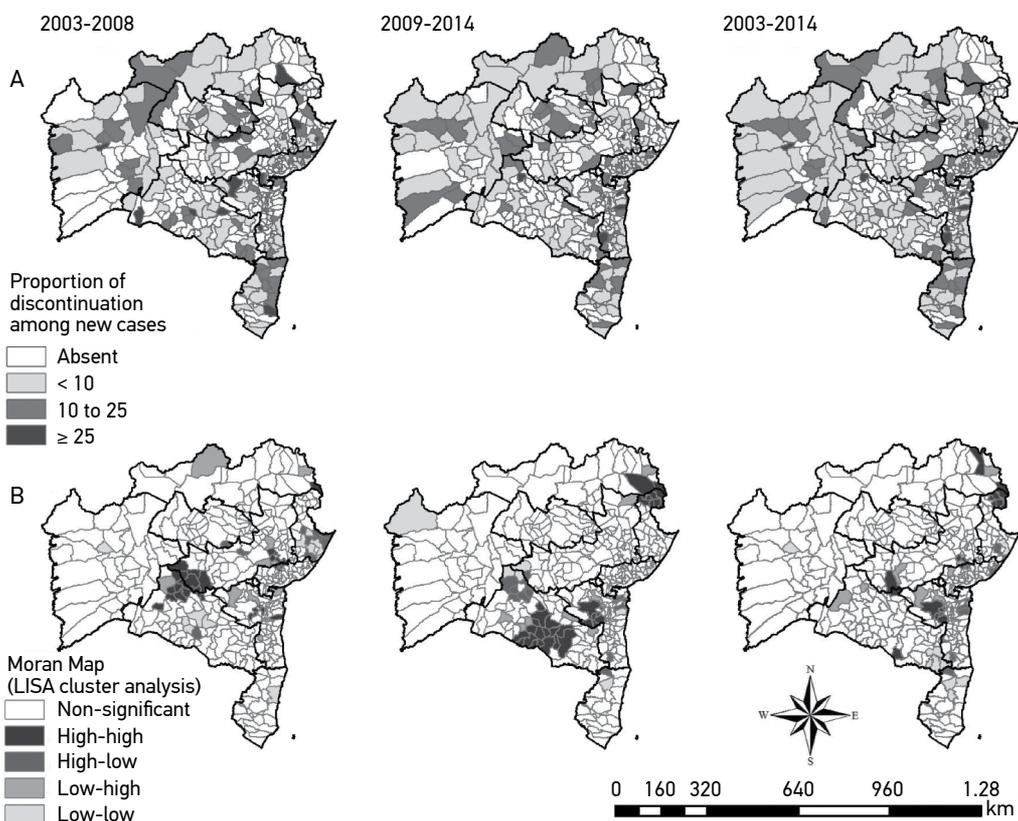

Figure 2. Spatial distribution of the proportion of treatment discontinuation among new leprosy cases diagnosed in the cohort years according to (A) municipality and spatial autocorrelation and (B) local Moran. Bahia, 2003–2008, 2009–2014, and 2003–2014.

From 2001 to 2008, 45.5% (n=190) of municipalities reported cases with G2D at the time of diagnosis. Out of them, 88 (46.3%) notified 10% or more of the new cases evaluated. These municipalities are located in all health districts of the state, particularly in the southwest and south districts. Between 2009 and 2014, the number of municipalities with a high incidence of G2D at diagnosis increased, reaching 55.3% (n=230) of the total, concentrated mainly in the west, north, and far-south districts (Figure 5A).

The local Moran analysis revealed areas of spatial correlation in the southwest, north, and northeast districts, indicating municipalities with high detection rates of cases with G2D surrounded by municipalities in the same situation. We identified a reduction in these areas of autocorrelation in the following period and the emergence of new clusters in municipalities of the east-central district, with persistence in the southwest of the state throughout the period (Figure 5B).
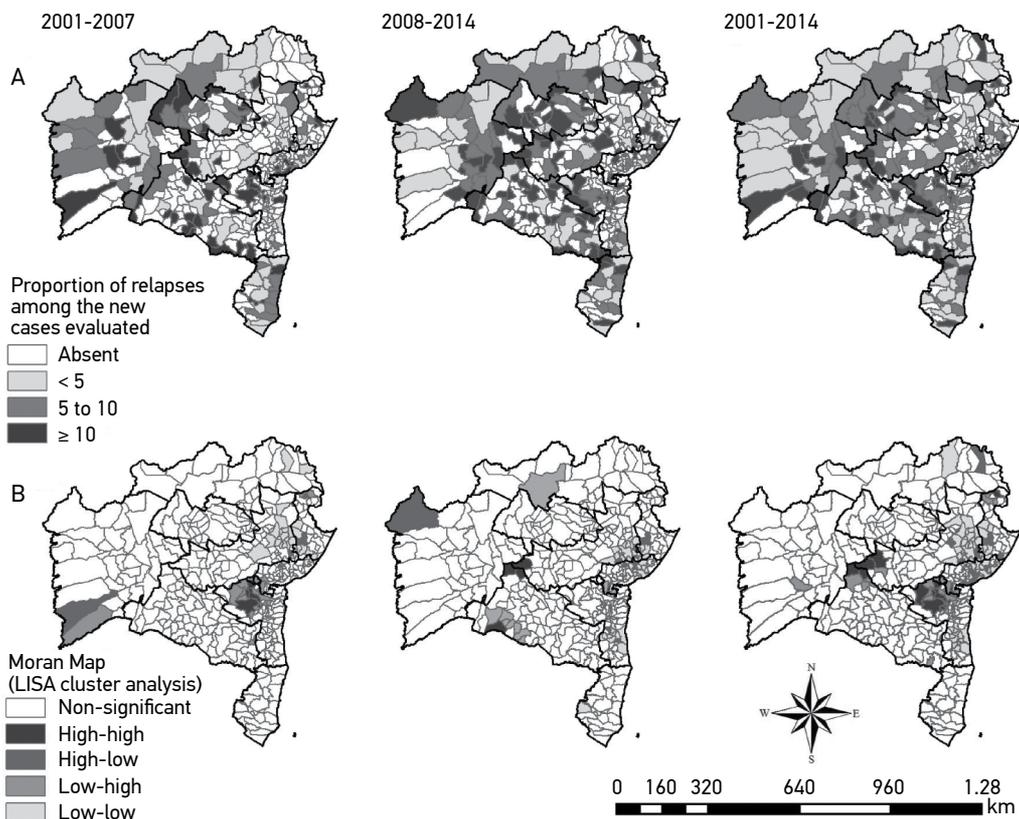

Figure 3. Spatial distribution of the proportion of leprosy relapse among the cases notified in the year according to (A) municipality and spatial autocorrelation and (B) local Moran. Bahia, 2001–2007, 2008–2014 and 2001–2014..

DISCUSSION

Most municipalities in the state of Bahia showed poor performance in leprosy control indicators, with stability or relative worsening in operational patterns over 14 years. We found poor or regular performances regarding the quality of care and follow-up provided until the cure, in addition to a growing number of municipalities with incidence of relapse. The performance of the disability grading at the time of diagnosis was regular, while the evaluation of the G2D indicator revealed low or average effectiveness of timely detection in a significant number of municipalities. Although not the object of this study, the integrated analysis of operational indicators points to potential endemic regions hidden in Bahia.

In addition to the considerable detection rate of new leprosy cases found in Bahia[12,15], the fact that most municipalities in this study presented a high percentage of patients who were
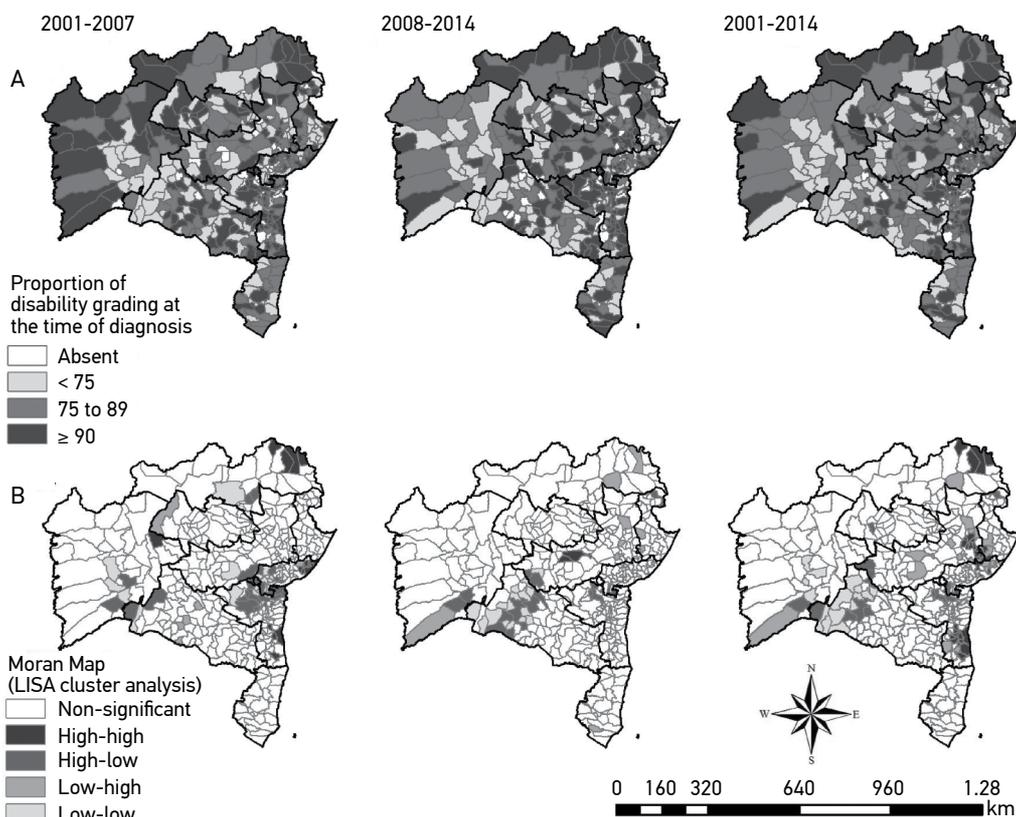

Figure 4. Spatial distribution of the proportion of new leprosy cases with disability graded at the time of diagnosis according to (A) municipality and spatial autocorrelation and (B) local Moran for new leprosy cases without disability graded at the time of diagnosis. Bahia, 2001–2007, 2008–2014 and 2001–2014.

not discharged for cure in the recommended period[4] increases the possibility of sources of infection that could be drug resistant[1,16,17]. This situation indicates significant care and surveillance failures and may contribute to physical impairment and stigma[1,15-17]. A study conducted in Bahia identified areas with a higher burden of the disease, particularly in the far-south, north, and west districts[15]. Some health districts show lower detection coefficients of new cases as well as unsatisfactory performance in the number of individuals discharged as cured, making its control even more complex[12].

In Brazil, even after the introduction of MDT, approximately 20% of patients were not cured in the cohort of 2017. This percentage is similar to that found in the state of Bahia, which holds the third-to-last place regarding performance in the Northeast Region[18]. São Luís do Maranhão faces an even more critical situation, considering that out of the 183 cases monitored, only 67.7% were discharged as cured[19]. Given the efficacy and effectiveness of
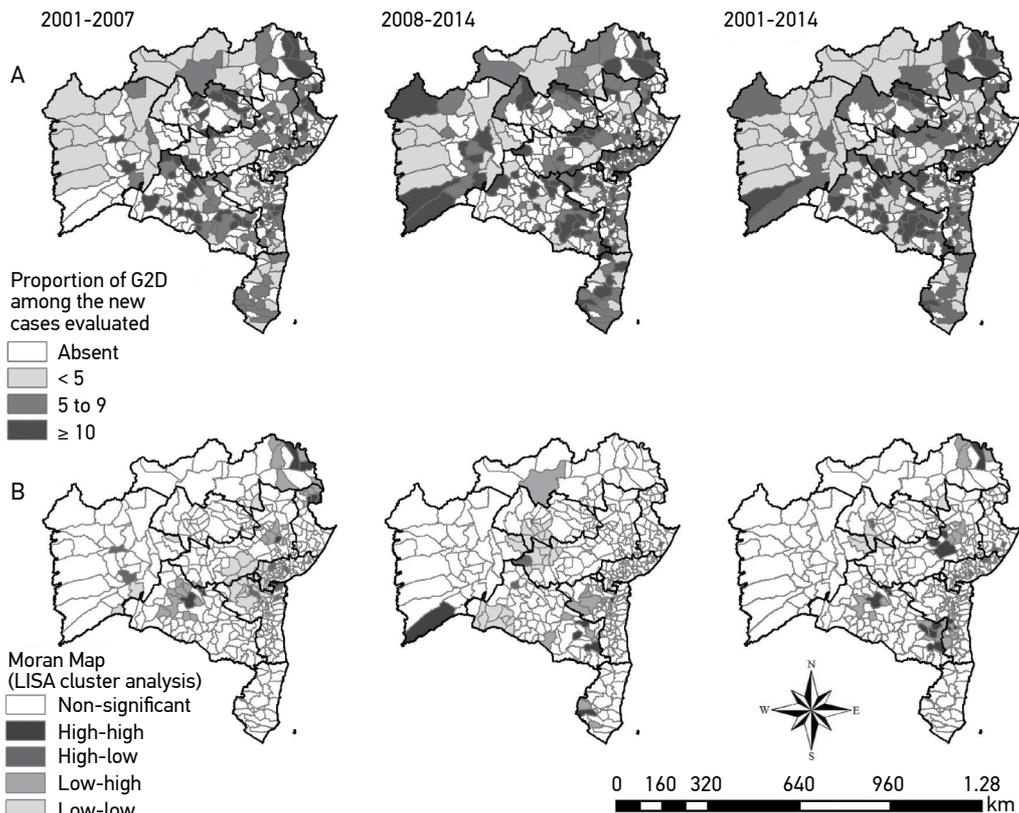

Figure 5. Spatial distribution of the proportion of new leprosy cases with grade 2 disability (G2D) at the time of diagnosis among new cases detected and evaluated according to (A) municipality and spatial autocorrelation and (B) local Moran. Bahia, 2001–2007, 2008–2014 and 2001–2014.

MDT, failures related to errors of classification and prescription, treatment irregularity, and MDT discontinuation might be associated with the lack of cure[20].

Also, the notification of treatment discontinuation is a reality in more than 30% of municipalities in Bahia, and small clusters of municipalities have emerged in areas with lower detection rates of new cases in the state[12,15]. Therefore, assessment studies addressing determining and conditioning factors to identify possible reasons for patients to discontinue their treatment should be encouraged. The absence of symptoms in early stages, the non-acceptance of the disease, the belief in religious cure, and the shame related to the monthly visits to health units were identified as causes for treatment discontinuation in the Brazilian Northeast[20]. In highly endemic areas in the state of Tocantins, treatment discontinuation was associated with males, illiterate individuals, those with multibacillary disease, who have difficulties in the route from home to the health service, with one or two people per household, under other treatments before the MDT, and who do not believe in cure[21]. Counseling at diagnosis can be a strategy to strengthen bonds, create spaces for listening, promote health education, and adopt behaviors to overcome vulnerability[22-24].

The number of municipalities with incidence of relapse is high, with growth among those that reported more than 10% of this type of case. We underline that its incidence is distributed in the nine health districts[15]. Thus, reviewing diagnostic criteria, discussing new MDT regimens for cases previously treated, and qualifying the follow-up are necessary[4,22].

Leprosy relapse is a complex subject, and its notification should follow the MoH guidelines[4]. In 2015, Brazil was responsible for 42.8% (n=1,452) of all cases of relapse reported to the World Health Organization (WHO)[25]. WHO estimates a risk of incidence of 1.1% after implementation of MDT for paucibacillary cases and 0.8% for multibacillary ones. In a study conducted in the state of Mato Grosso, 64.7% of municipalities have notified cases of relapse, corresponding to 6–20% of all cases. The predictive factors found for relapse are related to housing conditions, lifestyle habits, organization of health services, clinical forms, and treatment regimen[26]. Another study conducted in Bahia revealed an increasing trend in relapse among males[27]. In contrast, multicenter research that investigated drug resistance in five Brazilian states showed that this event has low magnitude and is present in old cases, with a history of institutionalization and subjected to long-term monotherapy regimens with irregular use[28].

In addition to qualifying health care, prioritizing the epidemiological investigation of cases of relapse following the criteria recommended by the MoH[4] is crucial. This is a key measure, considering the importance of differentiating therapy deficiency, therapeutic failure, and leprosy reactions, commonly mistaken in health services[4,28,29]. The diagnosis of relapse should be confirmed in reference centers for leprosy[4], but most municipalities in Bahia do not have proper access to specialized teams in their health districts.

We emphasize the importance of discussing issues related to professional health training, with the development of permanent education procedures and effective policies in health education for neglected events. These actions can positively affect the construction

of effective care networks, enabling integrated care, improving cure rates, and reducing treatment discontinuation and relapses[1,4,28].

The number of municipalities with regular or poor performance in the disability grading indicator was high. Similarly, most municipalities notified cases with G2D at the time of diagnosis, which increased over time, even in highly endemic areas, such as the west, north, and far-south districts of the state[4,12,15]. This scenario indicates late diagnosis and weaknesses in the development of actions to promote self-care and physical rehabilitation[4].

National data show that Bahia has the fifth worse performance in disability grading in the country[18], contrary to results of a study carried out in Londrina, Foz do Iguaçu, and Curitiba, whose percentages of disability grading at diagnosis were above 90%[30]. In addition to expanding the access to disability grading in the municipalities of Bahia, the quality of the examination and its proper notification to SINAN should be improved, since the proportion of individuals with disability graded and reported at the time of discharge is minimal. This finding also leads to the need for new studies, particularly operational ones, for a better understanding of the care process established, even in case of association of leprosy with other debilitating diseases and those of chronic evolution[1,11,31].

This study has potential limitations for being strictly based on secondary data. For instance, the low completeness of SINAN records regarding disability grading at the time of discharge prevented us from analyzing these data. We acknowledge the possibility of errors concerning the diagnosis of relapse by health services and the need for further research. This study did not find investigations on relapse in the state. However, both as to relapse and other indicators, the use of an extensive state historical series minimizes the effects related to non-completeness and inconsistency.

CONCLUSION

Bahia has a significant number of municipalities with poor or regular patterns of indicators for cure, relapse, GIF evaluation at diagnosis, and cases with GIF at the time of leprosy diagnosis. We underline the need to expand and qualify the surveillance and care provided by health services, taking into account the endemic maintenance in the state as to parameters of high endemicity. This scenario indicates the persistence of leprosy as a significant public health issue in Bahia in the coming years if the current operational control does not change.

This study revealed the existence of municipalities with poor patterns of operational indicators in all nine health districts, confirming the magnitude of the challenge for one of the largest Brazilian states. Thus, regional and/or municipal research can contribute to identifying weaknesses, especially with respect to primary health care, the implementation of leprosy control actions, and integrated care to individuals, families, and communities. In addition, we highlight the importance of strengthening SUS and tackling vulnerabilities with intersectoral actions to control leprosy.

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