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ORIGINAL ARTICLE

SPATIAL AND SPATIOTEMPORAL DISTRIBUTION OF *Schistosoma mansoni* CASES IN THE STATE OF ALAGOAS, BRAZIL

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ABSTRACT

This study aimed to analyze the occurrence of *Schistosoma mansoni* in the State of Alagoas, Brazil, over space and time from 2012 to 2021. An ecological, quantitative, and spatiotemporal study was conducted using secondary data from the Schistosomiasis Control and Surveillance System (SISPCE/DATASUS). Data were aggregated by health regions (HR) and municipalities. During the study period, 1,466,100 examinations were performed, and 73,325 cases of schistosomiasis were diagnosed. Spatial analysis showed that 20 municipalities had moderate positivity rates (5.1 – 15.0%), and one municipality had a high positivity rate above 15.1%. Global Moran's Index indicated significant positive spatial autocorrelation (I= 0.52308; *p*= 0.001). The Local Moran's Index identified 16 municipalities with a high-high pattern, distributed across the 1st, 2nd, 3rd, 4th, and 6th HR. The space-time scan analysis revealed two significant risk clusters. The primary cluster comprised 50 municipalities, predominantly located in the 3rd, 4th, and 6th HR, showing a relative risk (RR) of 11.06 (*p*< 0.001). The proportion of infections classified as severe was below 1%. These findings highlight the persistence of high-risk areas for schistosomiasis in Alagoas, concentrated in specific health regions, and are aligned with WHO goals to eliminate schistosomiasis as a public health problem.

KEY WORDS: Epidemiology; neglected diseases; public health; *Schistosoma mansoni*.

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INTRODUCTION

Schistosomiasis mansoni is a neglected tropical disease influenced by socioeconomic, structural, cultural, and political factors. The infection is caused by the digenetic trematode *Schistosoma mansoni*. *S. mansoni* is an obligate endoparasite requiring an invertebrate intermediate host – freshwater snails of the genus *Biomphalaria* – and definitive hosts, such as humans and other mammals, to complete its life cycle (Colley et al., 2014). Globally, schistosomiasis affects more than a quarter of countries and is geographically limited to tropical regions. In the Americas, *S. mansoni* is endemic in Venezuela, the Caribbean Islands, Suriname, and Brazil, where it is present in 19 Brazilian States (Brasil, 2014).

Estimates indicate that 1.5 million people in Brazil are infected, with approximately 42.9 million at high risk of infection (Brasil, 2022). Notably, 72% of cases occur in the Northeast region of the country (Paz et al., 2020), with the States of Sergipe, Alagoas, and Pernambuco showing the highest mortality and positivity rates (Martins-Melo et al., 2016; Paz et al., 2020; Alencar et al., 2024).

Schistosomiasis is a focal disease with areas of high prevalence, highlighting deep socioeconomic and health disparities (Rollemberg et al., 2015). It primarily affects populations facing economic and social vulnerability due to poor housing conditions and environmental infrastructure, inadequate sanitation, lack of hygiene, and low educational attainment (Bethony et al., 2006; Rollemberg et al., 2011).

Alagoas, endemic for the disease, ranks second to last in Brazil's Human Development Index (HDI), with a value of 0.684, ahead only of Maranhão, placing it 26th among Brazilian States (IBGE, 2022). This indicator reflects conditions that foster ongoing schistosomiasis transmission.

Therefore, identifying potential transmission hotspots is essential for planning public health policies of an educational and prophylactic nature, and aimed at health promotion, especially when articulated through intersectoral collaboration. This is particularly important in historically endemic regions, given the significant health impacts of the disease, which, if left untreated, can result in substantial morbidity and even mortality (McManus et al., 2018).

Hence, integrating epidemiological surveillance, basic sanitation, health education, and social policies is essential to strengthen control measures and mitigate the risks associated with the disease.

Thus, monitoring schistosomiasis positivity rates, along with their spatial and spatiotemporal patterns, is crucial given the endemicity scenario in the State. Such surveillance helps elucidate the current complexity of the public health issue and supports the evaluation of future trends (Cardoso et al., 2020). This study aimed to analyze the spatial and spatiotemporal occurrence of schistosomiasis mansoni in the State of Alagoas, Brazil, from 2012 to 2021.

MATERIAL AND METHODS

Study Design and Study Area

This is an ecological study with a quantitative approach, encompassing all 102 municipalities in the State of Alagoas, Brazil (Figure 1AB). The State has an estimated population of 3,127,687 inhabitants, distributed over a territorial area of 27,843,295 km² (IBGE, 2022). It is administratively divided into 10 health regions (HR) (Figure 1B).

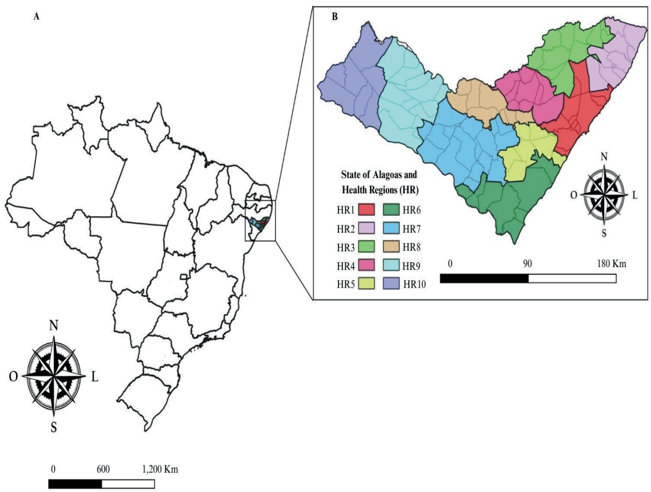

Figure 1. Map of the study area. (A) Map of Brazil highlighting the State of Alagoas. (B) Map of Alagoas divided by health regions (HR).

Data Collection and Sources

Data were obtained from the *Sistema de Informação do Programa de Controle da Esquistossomose* (SISPCE) and organized in Google Sheets® spreadsheets. All positive cases and diagnostic tests for *S. mansoni* reported between 2012 and 2021 were considered, aggregated by health region and municipality in Alagoas. These data were retrieved from SISPCE records available through the *Departamento de Informática do Sistema Único de Saúde* (DATASUS) platform (http://www2.datasus.gov.br/) under the Brazilian Ministry of Health. The municipal population data for Alagoas from 2012 to 2021 were obtained from the Brazilian Institute of Geography and Statistics (*Instituto Brasileiro de Geografia e Estatística* - IBGE) based on official intercensal estimates and were used for spatial analysis and map generation. We also collected cartographic shapefiles of the State, its municipalities, and HRs from the IBGE's Geographic Projection System in latitude/longitude coordinates based on the Geodetic Reference System (SIRGAS-2000).

Data Processing and Aggregation

The study used secondary, aggregated, and anonymized data on positive cases of *S. mansoni*, obtained from the Schistosomiasis Control and Surveillance System (SISPCE/DATASUS) and generated by the Schistosomiasis Control Program (*Programa de Controle da Esquistossomose* – PCE), through routine parasitological surveys conducted by municipal health services. Stool samples were examined using the Kato-Katz technique, with two slides per individual, following the recommendations of the World Health Organization (WHO). Individuals with at least one egg detected in the stool examination were considered positive cases.

The positivity rate for schistosomiasis mansoni in Alagoas was the primary variable of the study, calculated at the municipal level based on the number of positive cases. This variable was used in both the spatial and temporal analyses of the State, municipalities, and health regions studied. The positivity rate (PR) of *S. mansoni* infection was calculated as the number of positive individuals divided by the number of tests performed, multiplied by 100.

To better understand the disease dynamics in Alagoas, we also conducted a descriptive analysis of parasite burden in infected individuals. The intensity of infection was expressed as the number of schistosome eggs per gram of feces (EPG) and calculated as the total number of eggs among positives divided by the number of individuals examined, multiplied by 100.

The infection intensity was determined according to the Brazilian Ministry of Health guidelines for the Brazilian Schistosomiasis Control Program (PCE) considering the amount of EPG, as follows: light infection of 1 to 4 eggs in the feces and EPG up to 99, moderate infection of 5 to 16 eggs in the feces and EPG between 100–399, and heavy infection of >17 eggs in the feces and EPG >400.

Infection Percentage (IP) estimation:

Light-IP: (Number of individuals with 1–4 eggs on the slide / Total number of individuals examined) × 100

Moderate-IP: (Number of individuals with 5–16 eggs on the slide / Total number of individuals examined) × 100

Heavy-IP: (Number of individuals with ≥ 17 eggs on the slide / Total number of individuals examined) × 100

Spatial Distribution

After calculating the crude positivity rate, rate smoothing was performed using the local Empirical Bayesian model in TerraView® software, version 4.2.2. This method allows for the correction of potential data inconsistencies and random case occurrences, providing more stable positivity rate estimates. Thematic maps were generated to visualize the spatial distribution of both crude and smoothed positivity rates, classified according to the Ministry of Health as follows: low (0.1–5.0), moderate (5.1–15.0), and high (>15.0).

Spatial Autocorrelation Analysis

Spatial autocorrelation was performed to determine the presence of positive or negative spatial correlation using the Global Moran's Index (GMI). This index measures the spatial correlation of a variable with itself, ranging from -1 to +1, indicating whether the correlation is positive or negative and whether there are areas of similar spatial patterns. A positive value with $p < 0.05$ indicates positive spatial autocorrelation and suggests clusters of high infection risk for schistosomiasis. Values near zero indicate spatial randomness (Anselin, 1995).

The Local Moran's Index (LISA – Local Indicators of Spatial Association) was applied to identify clusters of similar values (high-high, low-low) and transition zones (high-low, low-high) (Anselin, 1995). All spatial distribution maps were created using the free software QGIS, version 3.32.3 (QGIS Development Team; Open Source Geospatial Foundation Project).

Retrospective Spatial and Spatiotemporal Scan Analysis

To identify spatiotemporal risk clusters, we conducted retrospective scan statistics using SaTScan software, version 9.6. This tool detects clusters using the Log Likelihood Ratio (LLR) test, represented by maps and tables (using QGIS version 3.32.3) to construct thematic maps. In this analysis, we considered the positivity rate for the disease, calculated as previously described, for municipalities with PCE activity. Non-endemic municipalities without PCE need to have a population for this respective analysis; therefore, population estimates for the municipalities were also used.

For each identified cluster, the relative risk (RR) of schistosomiasis compared to neighboring municipalities was calculated, along with the corresponding *p*-values. This standardization allows comparisons between different areas by minimizing the effects of varying population sizes and depicting the intensity of disease occurrence related to all study regions. We applied Kulldorff's (1997) retrospective space-time scan statistics using the Poisson distribution model with the following parameters: time aggregation unit of one year, with a maximum spatial cluster size of 25% of the at-risk population, and a maximum temporal cluster size of 50% of the study period.

RESULTS

During the study period, the PCE in Alagoas performed 1,466,100 examinations, of which 73,325 tested positives for *S. mansoni* (Table 1). The highest number of positive cases was recorded in 2012 and 2013 (11,384 and 12,652 cases, respectively). From 2014 onward, a continuous decrease in positive cases was observed (Table 1). Regarding the distribution by HR, the 3rd and 4th HRs presented the highest number of positive cases throughout the study period, with 21,526 and 20,370 cases, respectively (Table 1).

The results regarding positive rates by health regions in Alagoas revealed low rates, mostly below 5% over the entire study period. However, the 3rd and 4th HRs recorded the highest positivity rates for schistosomiasis mansoni, with 8.1% and 6.9%, respectively (Table 1).

Regarding infection intensity based on EPG for *S. mansoni*, light infections were predominant across the State (light: 3.7%, moderate: 1.0%, heavy: 0.3%). Once again, the 3rd and 4th HRs stood out with the highest indicators of infection intensity in their populations. Light infection rates were 6.1% and 4.9%; moderate infections reached 1.7% and 1.6%; and heavy infections were observed at 0.4% and 0.5%, respectively, considering the period studied (Table 2).

Table 1. Tests and positivity by health regions in the State of Alagoas, according to the Schistosomiasis Control Program (PCE).

Health Regions	Variables	2012	2013	2014	2015	2016	2017	2018	2019	2020	2021	Total
	Exame	17,633	28,656	27,644	30,676	23,503	27,402	23,895	24,812	12,062	17,179	233,462
1st HR	Positive	1,056	1,528	1,001	1,018	981	918	642	722	241	410	8,517
	TP %	5.9	5.3	3.6	3.3	4.1	3.3	2.6	2.9	2.0	2.3	3.6
	Exame	9,772	9,855	7,789	5,760	5,395	10,145	12,554	12,672	3,235	10,384	87,561
2nd HR	Positive	479	317	299	294	212	398	402	393	80	258	3,132
	TP %	4.9	3.2	3.8	5.1	3.9	3.9	3.2	3.1	2.4	2.4	3.5
	Exame	34,958	33,428	31,933	27,803	22,190	24,508	28,169	28,076	10,408	23,797	265,270
3rd HR	Positive	3,290	4,154	2,866	2,240	1,874	2,066	1,577	1,830	627	1,002	21,526
	TP %	9.4	12.4	8.9	8.1	8.4	8.4	5.6	6.5	6.0	4.2	8.1
	Exame	38,706	36,851	35,543	34,835	29,157	29,664	28,594	31,036	10,129	20,450	294,965
4th HR	Positive	3,267	3,679	2,973	2,310	1,981	1,735	1,446	1,385	451	1,143	20,370
	TP %	8.4	9.9	8.3	6.6	6.7	5.8	5.1	4.4	4.4	5.5	6.9

	Exame	11,154	13,034	14,737	17,166	14,180	14,617	16,107	15,609	11,180	10,715	138,499
5th HR	Positive	333	401	240	273	309	264	336	187	165	205	2 713
	TP %	2.9	3.1	1.6	1.5	2.1	1.8	2.1	1.2	1.4	1.9	1.9
	Exame	21,894	20,980	17,211	16,114	10,066	19,381	19,563	20,488	15,286	18,910	179,893
6th HR	Positive	1,707	1,421	1,187	909	412	1,091	1,018	800	573	716	9,834
	TP %	7.8	6.7	6.9	5.6	4.1	5. 6	5.2	3.9	3.7	3.7	5.4
	Exame	15,351	14,025	15,962	16,037	15,654	12,590	14,522	15,022	10,454	8,824	138,441
7th HR	Positive	671	724	652	602	545	316	312	207	145	53	4,227
	TP %	4.3	5.1	4.1	3.7	3.4	2.5	2.1	1.3	1.3	0.6	3.1
	Exame	17,330	17,261	11,814	10,415	10,952	9,935	13,205	15,055	10,544	11,498	128,009
8th HR	Positive	581	428	557	319	193	242	247	235	71	133	3,006
	TP %	3.3	2.4	4.7	3.1	1.7	2.4	1.8	1.5	0.6	1.1	2.3
	Exame	166,798	174,090	162,633	158,806	131,097	148,242	156,609	162,770	83,298	121,757	1,466,100
Total	Positive	11 384	12 652	9 775	7 965	6 507	7 030	5 980	5 759	2 353	3 920	73 325
	TP %	6.8	7.2	6.0	5.0	4.9	4.7	3.8	3.5	2.8	3.2	5.0

Classifications follow the Brazilian Ministry of Health: Low (0.1–5.0%), Moderate (5.1–15.0%), High (>15.0%). HR = Health Region; PR = Positivity Rate.

Table 2. Percentage of intensity of infection by *Schistosoma mansoni*, in the State of Alagoas and health regions, by positivity range.

Alagoas	Number of Eggs				%Total		
HR	1-4	5-16	>17	Exams	mild	moderate	intense
1st HR	6,500	1,589	428	233,462	2.8	0.7	0.2
2nd HR	2,373	621	138	87,561	2.7	0.7	0.2
3rd HR	16,109	4,387	1,030	265,270	6.1	1.7	0.4
4th HR	14,370	4,672	1,328	294,965	4.9	1.6	0.5
5th HR	2,167	442	104	138,499	1.6	0.3	0.1
6th HR	7,883	1,652	299	179,893	4.4	0.9	0.2
7th HR	3,001	1,124	102	138,441	2.2	0.8	0.1
8th HR	2,013	746	247	128,009	1.6	0.6	0.2
Total	54,416	15,233	3,676	1,466,100	3.7	1.0	0.3

HR = Health Region.

To identify risk areas for *S. mansoni* infection, we analyzed the spatial and spatiotemporal distribution of the disease's positivity rate in the State (Figure 2A). The results revealed a heterogeneous distribution across the endemic HRs, with most areas showing positivity rates below 5.0%, indicating low endemicity.

Our analysis also identified 20 municipalities with moderate endemicity (positivity rates between 5.1% and 15.0%). Only one municipality (Branquinha) exhibited a positivity rate above 15.0%, thus classified as highly endemic. When applying the local empirical Bayesian smoothing method, the spatial patterns were only slightly modified, with no significant differences in the overall positivity rates across the State (Figures 2AB).

Furthermore, the analysis revealed a positive spatial autocorrelation (GMI = 0.52308; $p = 0.001$). The LISA index identified 16 municipalities with a high-high (Q1) cluster pattern, indicating high risk for the disease, located in the 1st, 2nd, 3rd, 4th, and 6th HRs. Another 27 municipalities in the 7th, 8th, 9th, and 10th HRs exhibited a low-low (Q2) pattern, indicating low-risk areas – particularly in the 9th and 10th HRs, which are not endemic for the disease. Three municipalities in the 3rd and 8th HRs showed a low-high (Q3) pattern, suggesting a transitional risk (Figure 2C).

Lastly, retrospective space-time scan statistics identified two high-risk clusters. The primary cluster encompassed 50 municipalities across all endemic areas for schistosomiasis of the State, with an RR of 11.06 ($p < 0.001$) and a log-likelihood ratio of 46,895.10 (Figure 2D; Table 3). The secondary cluster was in one municipality within the 7th HR (Feira Grande), with an RR of 8.56 ($p < 0.001$) and a log-likelihood ratio of 674.44 (Figure 2D; Table 3).

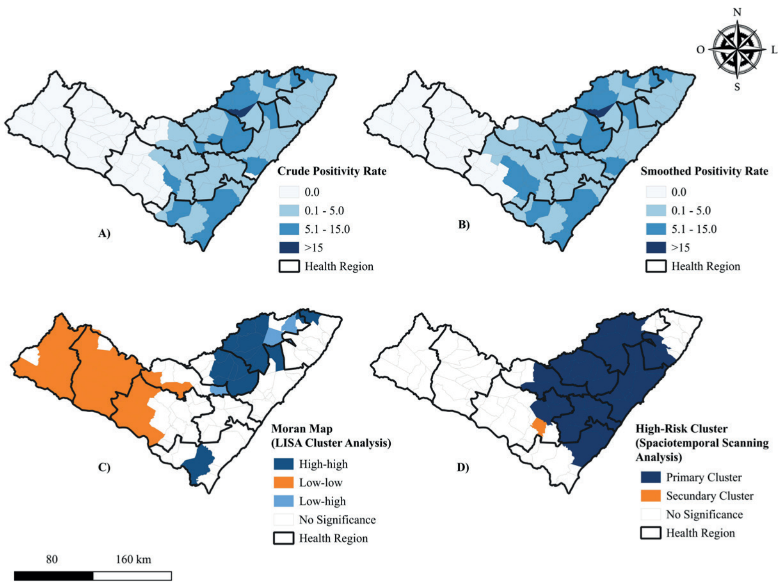

Figure 2. Map of spatial and spatiotemporal statistical analysis of the positivity rate for *Schistosoma mansoni* in Alagoas. (A) Spatial distribution of schistosomiasis mansoni, according to the crude positivity rate; (B) Spatial distribution of schistosomiasis mansoni, according to the smoothed positivity rate; (C) Spatial distribution of spatiotemporal autocorrelation according to Moran's index; (D) Spatiotemporal scan analysis.

Table 3. Spatiotemporal groupings of annual rates of positive cases for *Schistosomiasis mansoni* in the State of Alagoas.

Cluster	Mun.	Períod	Number of Cases	New Cases Expected	PR/100 000 hab.	RR	LLR	*p*-value
1	50	2012 a 2016	41880	7883.39	4610.3	11.06	46895.101193	< 0.001
2	1	2012 a 2015	535	62.93	7377.4	8.56	674.447524	< 0.001

Clusters were identified using retrospective space–time scan statistics (Poisson model) in SaTScan v9.6. The study analyzed data from 2012 to 2021; however, the detected clusters were concentrated in the periods 2012–2016 and 2012–2015. RR = Relative Risk; LLR = Log Likelihood Ratio.

DISCUSSION

Over the ten-year study period (2012 to 2021), the State of Alagoas recorded 73,325 positive cases of schistosomiasis mansoni, according to data from SISPCE. During this period, a slight but progressive reduction was observed in both the number of positive cases and the positivity rate of disease in the last five years. We also found that most *S. mansoni* infections in the State presented low infection intensity, with 3.7% of the cases classified as light infections. This pattern reflects a lower parasitic burden, possibly due to reduced exposure to the intermediate host and partial success of preventive and control measures. Regarding the proportion of more severe infections within these municipalities, the State reduced the number of infections to less than 1% during the years of study.

These findings indicate a potential positive impact of the actions of the PCE in the State. The observed reduction aligns with one of the main goals set by the World Health Organization (WHO), which aims to reduce schistosomiasis positivity rates to levels that render the disease no longer a significant public health problem by 2030 (WHO, 2022).

However, according to Alencar et al. (2024), with the implementation of surveillance and diagnostic actions for the disease, as well as preventive chemotherapy in endemic areas, a more substantial decrease in the number of positive cases and positivity rates would be expected, rather than the subtle decline observed. This suggests that there may be gaps in PCE coverage in certain endemic areas of the State. Additionally, it is noteworthy that the reduction in schistosomiasis cases and positivity rates during 2020 and 2021 may be attributed to the interruption of PCE activities in Alagoas due to other public health emergencies, such as the COVID-19 pandemic. Consequently, this may have affected data accuracy regarding the actual extent of the disease in the State, as reported by Dantas et al. (2023), who identified significant impacts of the pandemic on schistosomiasis surveillance and control activities in endemic areas of Northeastern Brazil, including the suspension of parasitological surveys and mass treatment actions.

Importantly, spatial analysis methods have been used to predict schistosomiasis occurrence (Bailey and Gatrell, 1995). The spatial and spatiotemporal distribution of positivity rates across the State revealed significant heterogeneity among endemic Health Regions. Although most areas showed positivity rates below 5%, indicating low endemicity, 20 municipalities had moderate positivity rates (5.1% to 15.0%), and one municipality exhibited a high positivity rate (>15.0%).

This uneven distribution may be associated with several internal and external social factors, as emphasized by Coura-Filho et al. (1995) and Tibiriçá et al. (2011), including local socioeconomic conditions, access to healthcare services, the effectiveness of schistosomiasis control activities, and the implementation of health education initiatives. It may also reflect current guidelines from the Ministry of Health, which recommend that only populations from localities with positivity rates above 25% receive complete community treatment, whereas localities with rates below 25% treat only confirmed cases and household contacts, potentially minimizing the infection rate (Brasil, 2024) and contributing to the maintenance of low to moderate positivity rates over time.

The positive spatial autocorrelation identified by the GMI reinforces the existence of high-risk areas located near each other, suggesting a tendency for the disease to cluster geographically over time. Compared to the findings of Santos et al. (2020), Alagoas has reduced its number of high-risk municipalities from 22 to 16, characterizing them as high-risk areas. In contrast, 27 municipalities were identified as low-low clusters, indicating low-risk or non-endemic areas, fewer than previously reported.

These results are consistent with the findings of Tibiriçá et al. (2011), which showed that expanded healthcare coverage and the incorporation of new diagnostic and surveillance technologies by the PCE have led to improved detection of schistosomiasis and more effective control of endemic areas. However, these efforts have not been sufficient to eliminate the spatial expansion of schistosomiasis in the State, as the disease's focal nature is strongly influenced by environmental and socioeconomic determinants (Coutinho et al., 1992).

The spatiotemporal cluster analysis identified two high-risk clusters for schistosomiasis. The primary cluster encompassed 50 of the 70 endemic municipalities identified by Santos et al. (2020), with a significantly high relative risk (RR = 11.06; $p < 0.001$). The association of these high-risk areas with PCE coverage and adequate treatment of affected populations is critical for reducing, maintaining, or potentially increasing disease occurrence (Santos et al., 2020). However, the persistence of high-risk areas in the State appears to be related to cultural factors and the absence of effective public policies, such as limited regional coverage of preventive chemotherapy and diagnostic failures within the PCE (WHO, 2012; Silva-Moraes et al., 2019).

Our findings corroborate previous evidence from studies conducted in Northeast Brazil, where spatial and spatiotemporal analyses have revealed the persistence of schistosomiasis-related morbidity and mortality despite long-term control efforts (Silva et al., 2021; Silva et al., 2022).

Thus, mapping the spatial distribution of risk areas has provided valuable insights into the current situation of municipalities across the State. These findings offer scientific support for the detection and prevention of future cases and serve as a tool for planning and implementing strategies to improve health services and control schistosomiasis in Alagoas.

Accordingly, these findings are in line with the World Health Organization's (WHO) global strategy to eliminate schistosomiasis as a public health problem by 2030, aiming to reduce the number of severe infections in endemic areas to less than 1%. However, we emphasize that the data collected through SISPCE may present inconsistencies due to underreporting, data entry errors, and incomplete records, which are limitations of this study as it is based on secondary data. These factors may influence the final analysis of the actual epidemiological situation of schistosomiasis mansoni in Alagoas. Therefore, the study presented the spatial and spatiotemporal distribution of positive cases of schistosomiasis in the State, even with evidence of potential underreporting in certain areas.

The spatial analyses were used to identify possible high-risk areas, epidemiological clusters, and gaps in disease surveillance that warrant attention. Through spatial and spatiotemporal data, it was possible to define areas of elevated risk and limited health promotion coverage. Although positivity rates were heterogeneously distributed across the State, they were more predominant in municipalities with greater coverage by the PCE.

Our findings underscore the importance of maintaining continuous schistosomiasis control measures, particularly in endemic areas, and reinforce the need for a more geographically balanced approach to healthcare delivery and more effective disease notification and surveillance systems.

CONFLICT OF INTEREST

There is no conflict of interest to declare.

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