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Short Communication

American cutaneous leishmaniasis in a northeast Brazilian city: clinical and epidemiological features

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Abstract

Introduction: This study characterized the clinico-epidemiological profile of American cutaneous leishmaniasis (ACL) cases in Barbalha, Ceará State, Brazil. **Methods**: Medical records of 363 patients visiting Federal University of Cariri between 2009 and 2014 were analyzed. **Results**: ACL was more prevalent in men with low education level from rural zones. The main presentation was a single ulcer, mainly in the lower limbs, and 49.8% also presented lymphadenomegaly. The annual incidence ranged from 2.83 to 22.60 per 10,000 inhabitants. **Conclusions:** The rates observed in this study indicate the importance of additional research to contribute to the control of this endemic disease.

Keywords: American cutaneous leishmaniasis. Cutaneous leishmaniasis. Public health.

According to the World Health Organization (WHO), the epidemiology of cutaneous leishmaniasis in the Americas is complex, with wide variation in transmission cycles, hosts, vectors, clinical manifestations, and response to therapy, with multiple circulating *Leishmania* species[1].

Brazil is one of ten countries responsible for more than 70% of all cutaneous leishmaniasis cases[2]. The inoculation of parasites is responsible for cutaneous lesions in the entrance area[3].

Data analysis from DataSUS[4] showed that from 1980 to 2010, Ceará State had the higher number of cases of American cutaneous leishmaniasis (ACL) in the Northeast region. Additionally, one of the most affected areas in the State was the Cariri region.[5] Silva[6] showed that the highest incidence of leishmaniasis in Cariri occurred mainly in plateau areas near the foothills of the sierra, particularly the cities of Crato, Juazeiro do Norte, Jardim, and Barbalha.

Despite the high prevalence in Barbalha, there is a lack of clinico-epidemiological data regarding this region. Thus, this study aimed to determine the clinico-epidemiological profile of the ACL cases based on data from patients diagnosed at our service.

The data collection was performed at the Tropical Medicine Outpatient Clinic of the Federal University of Cariri, located in Barbalha. This city is within the Cariri region, in the Southern region of Ceará State, Brazil.

A retrospective and descriptive cross-sectional analysis was performed based on the medical records archived from January 1, 2009, to December 31, 2014. The inclusion criteria were a) patient from Barbalha; b) diagnosis of ACL; c) sufficient data about the case; and d) non-repeat patient. The data collection followed two standardized protocols. The first included identification data and socioeconomic variables, while the second included clinical data. In this study, the variable "size of lesions" was defined as the sum of the diameters of all lesions in each patient. The results of laboratory tests were also included in the second protocol. The techniques and procedures of parasitological diagnosis were performed as described previously[7].

ACL cases were confirmed clinically and epidemiologically. In addition, Montenegro's test was also used. The collected

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information was tabulated and processed in a Microsoft Office Excel® 2010 spreadsheet. These data were presented using descriptive statistics.

Information from the aforementioned outpatient clinic was compared with epidemiological indicators obtained from the State Secretariat of Health of Ceará[8]. This study abides by the Resolution N°466/2012 of the Brazilian Ministry of Health and was approved by the University's Ethics Committee under registration number 1.793.646.

Among the 363 included cases, 205 patients were male (56.47%) and 158 were female (43.53%), with an average age of 34.7 years (**Table 1**). The main occupations for both genders were farmers and students. Most patients (89.53%) were from rural zones. Other demographic data are shown in **Table 1**. The annual incidence of cases attended at our service ranged from 2.83/10,000 inhabitants in 2012 to 22.60/10,000 in 2010 (**Figure 1**). **Figure 1** also presents data from Brazil and three other countries (Peru, Colombia, and Paraguay) among the 12 high-burden ACL countries in 2014, according to WHO[2]. Nicaragua and Panama were also evaluated because they had the highest incidence rates in 2009–2014 based on data from PAHO[9].

Moreover, 316 cases included data on the type of lesion and 237 included data on the lesion size. The lesion types included papule in five patients (1.38%), pustule in nine patients (2.48%), ulcers in 292 patients (80.44%), scar in one patient (0.28%), nodule in one patient (0.28%), plaque form in one patient (0.28%), and mucocutaneous form in three patients (0.83%).

Analysis of the number of lesions revealed that single lesions (68.04%) prevailed over multiple lesions (27.0%). Four (1.10%) cases presented disseminated disease and it was not possible to identify the number of lesions in 3.85% of cases. Lymphadenomegaly was present in at least, one region of the body in almost 50% of the patients, most commonly in the inguinal lymph nodes. There were cases in which patients had no visible lesions, only lymphadenomegaly. Later, some developed ulcers which were observed and registered during follow-up (information provided by the staff of the service). Among the 304 patients (83.75%) with lesions on only one part of the body, the most affected body region was the lower limbs (164 patients, 45.18%). The other regions included the upper limbs 82 (22.59%), head 34 (9.37%), and trunk 24 (6.61%). There was one case with a lesion with an atypical presentation at the glans of the penis that measured 10 mm. The smallest lesion registered was 2 mm, while the largest affecting one single patient was 190.6 mm in diameter (based on the sum of diameters). The mean size of the lesions was 23.4 mm.

Regarding comorbidities, 52 patients (11.57%) had hypertension, nine (2.47%) had diabetes, and five (1.47%) had asthma. In contrast, 299 patients (82.37%) had no comorbidities.

Regarding the diagnostic methods, 199 (54.82%) cases were confirmed by laboratory tests. **Table 2** shows the results of the tests used, as well as the numbers of patients who tested negative or had a combination of positive results. A total of 120 cases were positive for *Leishmania (Viannia) braziliensis* by immunohistochemistry.

According to Americas data from PAHO, 69% of the cases were men, and approximately 50% were aged between 20 and 50 years[9]. In line with these data, our study showed a slight predominance of male subjects (56.47%), with ages ranging from 19 to 40 years. The prevalent labor activity was farming, reported by almost half of the male patients, similar to the reports from Bustos et al.[10] and Teles et al.[11].

The transmission of this disease is related to human activity in the vectors' habitat. This pattern also occurs in Barbalha, in which mainly men are affected, as those in the Cariri region usually work outdoors in fields within the vectors' habitat during the peak vector activity.

Teles et al.[11] reported that in addition to the prevalence of leishmaniasis in people living or working in rural areas, the vector is also adapting to peri-urban areas. Similar to Bustamante et al.[12], 10.18% of the patients in the present study did not live in rural areas, which might indicate the transmission of this disease outside these regions. However, we cannot exclude the possibility of cases caused by recreational or casual visits to rural areas.

In the present study, 198 (54.6%) patients were frequenting or had frequented the elementary school. The high rate of patients with low educational level suggests that this disease affects mainly people with low socioeconomic status, as has been reported in Argentina[10].

The clinical presentation of cutaneous leishmaniasis lesions can mimic several other disorders, thus confounding physicians and leading to a delay in precise diagnosis[13]. In the present study, the atypical presentations included a plaque form, a scar lesion, and a penile lesion. Gontijo and Carvalho[3] described a great plurality of clinical conformations of ACL in an endemic area, which was also found in our sample. However, ulcers were the most prevalent lesion type.

Since the lower and upper limbs are generally the parts of the body most exposed to the vector due to the tropical weather in the Cariri region, they were also the most affected parts of the patients' body, followed by the head, mainly the cheeks. This finding demonstrates the necessity to develop methods to better protect these parts of the body from parasite inoculation.

According to the literature, simultaneous presentation of lymphadenopathy and cutaneous lesions are found in *Leishmania (Viannia) braziliensis* infections[10]. Another study in Ceará reported that 77% of the patients presented with lymphadenopathy in addition to skin lesions and that *Leishmania braziliensis* should be considered in cases of unexplained lymphadenopathy in endemic areas[14]. In line with this report, the prevalence of lymphadenopathy in the present study was about 50% and all 120 cases submitted to immunohistochemical examination were positive for *L. (Viannia) braziliensis*.

Although the Brazilian Ministry of Health recommends diagnosis confirmation through laboratory methods before initiating treatment, this delay allows a pure clinico-epidemiological diagnosis, especially when the patient is from an endemic area[15]. In this study, 54.82% of the cases underwent at least one confirmatory test. Resource limitations

TABLE 1: Sociodemographic characteristics present in the analyzed records of the Tropical Medicine Ambulatory.

Characteristics	Female		Male		Mean
	n	%	n	%	
Age (years)					34.76
1–18	33	9.09	52	14.32	
19–40	71	19.55	87	23.96	
41–60	28	7.71	41	11.29	
>60	25	6.88	23	6.33	
Not reported	1	0.27	2	0.55	
Schooling					
Under age	8	2.20	4	1.10	
Illiterate	14	3.85	23	6.33	
Literate	0	0.00	1	0.27	
Elementary school	55	15.15	105	28.92	
High school	29	7.98	33	9.09	
Higher education	7	1.92	2	0.55	
Not reported	45	12.39	37	10.19	
Occupation					
Under age	8	2.20	4	1.10	
Student	35	9.64	56	15.43	
Farmer	38	10.47	87	23.97	
Retired	18	4.96	12	3.31	
Teacher	6	1.65	1	0.28	
Merchant	1	0.28	4	1.10	
Housewives	31	8.54	0	0.00	
Other	7	1.93	29	7.99	
Not reported	14	3.86	12	3.31	
Zone					
Urban	19	5.23	18	4.95	
Rural	139	38.29	186	51.23	
Not reported	0	0.00	1	0.27	
Time until firstconsultation (months)					
< 1	53	14.6	80	22.04	
1–3	91	25.07	110	30.3	
>3	8	2.2	6	1.65	
Not reported	6	1.65	9	2.48	

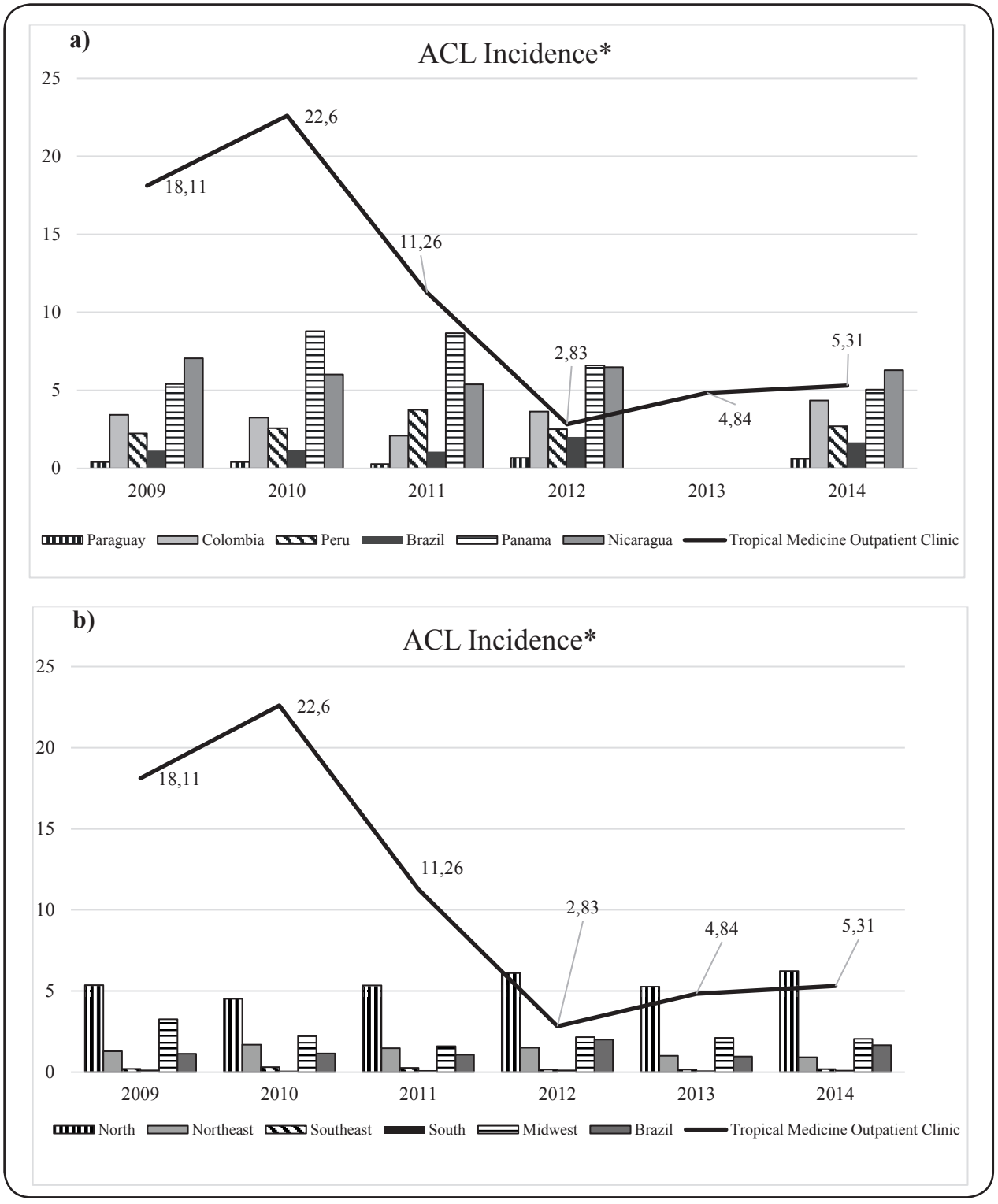

FIGURE 1: **Comparative data of ACL incidence rates from the Tropical Medicine Outpatient Clinic (Barbalha) with: a)** rates from Brazil and other Latin America Countries (Paraguay, Colombia, Peru, Brazil, Panama, and Nicaragua); **b)** and Brazilian geographic regions (North, Northeast, Southeast, South, and Midwest).
a. Country rates were calculated based on data from an epidemiological report of the Pan American Health Organization - PAHO (reference 9).
b. Data on Brazilian regions were obtained from DataSUS, Ministry of Health, Brazil.
*Incidence rate (cases/10,000 inhabitants).
**The PAHO report contained no data from 2013.

TABLE 2: Summary of the Diagnostic Methods performed at the Tropical Medicine Ambulatory of the Faculty of Medicine, Federal University of Cariri.

Diagnostic Methods	Positive (%)	Negative (%)	Discarded (%)	Not performed (%)	
Histopathological	99(27.3)	54(14.9)	-	210(57.9)	
Imprint	137(37.7)	7(2)	-	219(60.3)	
Montenegro test	168(46.3)	17(4.7)	-	178(49)	
Culture	80(22)	1(0.3)	44(12.1)	238(65.6)	
Immunohistochemistry	120(33.1)	0	-	243(66.9)	
Number of confirmatory tests in each patient	**0**	**1**	**2**	**3**	**4**
Number of cases (%)	164(45.2)	58(16)	45(12.4)	49(13.5)	47(12.9)

for routine laboratory examinations led to delayed diagnosis in the remaining cases, which were based mainly on clinico-epidemiological features and therapeutic response.

Based on WHO data[2], the percentage of diagnoses confirmed by laboratory tests in our service was less than those in other countries in 2014, including Paraguay, Colombia, and Peru which covered 100% of cases, and even Brazil (89%). From a continental perspective, our service presented values similar to those of Nicaragua and Mexico and greater than that of Argentina (approximately 20%) in 2014[9]. However, it is necessary to expand the use of confirmatory tests; although the diagnosis of ACL is a hard task, it is essential considering the toxicity profiles of the available drugs.

A lower incidence rate was observed in 2012 in our service during the study period, similar to the rates reported in Peru and Colombia but higher than those in Brazil and Paraguay[2]. In addition, in 2009–2011, the rates in Barbalha were higher than those in Nicaragua and Panama, the countries with the highest rates in that period[9]. It is worth noting that Barbalha registered case numbers higher than those in some Latin American countries, including Paraguay.

In summary, the main clinico-epidemiological features were the predominance in men aged 19–40 years from mainly rural areas, with a single ulcerated lesion generally less than 30mm in any part of the limbs. Half of the cases presented with lymphadenopathy. The absence of confirmatory laboratory tests in 45.2% of the cases along with the non-identification of *Leishmania* species in the majority of the samples were the major limitations of this study. However, the rate of non-confirmed cases by laboratory tests was less than those from other Latin American countries. Finally, the incidence rates showed the relevance of ACL in the Cariri region and indicate the need for additional research in order to better characterize this disease throughout the country, provide information to guide effective health public programs, and improve the management of this illness among clinicians working in regions with similar features.

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Conflict of Interest

The authors declare that there is no conflict of interest.

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