Original Article

Epidemiology and clinical aspects of tungiasis (sand flea infestation) in Alagoas State, Brazil

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Abstract

Background: Tungiasis (infestation with the sand flea *Tunga penetrans*) is common in resource-poor populations throughout Brazil. However, the epidemiological situation and the clinical aspects are not fully understood.

Methodology: To describe the prevalence and severity of tungiasis, associated pathology, as well as the seasonal variation in rural northeast Brazil, we performed two cross-sectional surveys: one in the rainy season, another in the dry season. Individuals were examined for the presence of tungiasis, number of lesions, symptoms and signs. In the rainy season, 88.6% (1,015/1,146), and in the dry season 91.1% (990/1,087) of the respective target populations were examined.

Results: The prevalence of tungiasis was 21.6% (95% confidence interval: 19.0-24.1) in the rainy season and 29.5% (26.6-32.3) in the dry season (p<0.0001). The highest prevalence was found in boys (5 – 14 years) in the dry season (48.3%; 40.6-56.0). Most lesions occurred on the feet (rainy season: 96.3%; dry season: 97.5%); a considerable number of individuals presented with lesions on the hands (6.9% and 5.1%, respectively). Common symptoms and signs were desquamation of the skin (57.5% in the rainy season; 44.5% in the dry season), hyperkeratosis (51.6% and 34.6%) and nail deformation (32.0% and 23.3%). Superinfection was present in 15.5% and 13.7% of cases, respectively. Severe pathology, such as deep fissures (10.5% and 9.3%), loss of toe-nails (5.5% and 2.4%) and difficulty walking (1.4% and 0.7%) occurred less commonly.

Conclusions: Tungiasis occurs to an important degree in Alagoas State, and prevalence and morbidity varies according to the season. Children were identified as a high-risk group.

Key Words: tungiasis, epidemiology, prevalence, morbidity, Brazil.

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Introduction

The tropical parasitic skin disease tungiasis is defined as an infestation with the sand flea *Tunga penetrans* (Linnaeus, 1758). The female flea penetrates into the epidermis and undergoes an extraordinary hypertrophy, eventually reaching the size of a pea.

Tungiasis is endemic in many communities in Latin America, from Mexico in the north to Argentina in the south, on several Caribbean islands, and in sub–Saharan Africa [1-7]. *T. penetrans* occurs in resource-poor communities in rural areas, in fishing villages along the coast, and in the slums of big cities. In poor communities in Brazil, Trinidad, Cameroon and Nigeria, prevalences ranged between 16% and 54% [4-6, 8-15]. In these settings the parasite load is high, and individuals harbour dozens or even hundreds of fleas [2,6,16].

Complications due to tungiasis are common in areas where constant re-infestation is the rule [1, 7,16]. Bacterial superinfection is often present [17]. Severe inflammation and deep fissures in the skin hinder individuals from walking normally. Sequels include deformation and loss of toenails, as well as deformation of digits [1,7,16]. Lesions may also be port of entry for *Clostridium tetani* [2,7,18,19].

In the State of Alagoas (northeast Brazil) anecdotal reports indicate the occurrence of tungiasis, but systematic studies assessing epidemiological and clinical characteristics have never been performed. To fill this gap, we performed a population-based study on tungiasis in a rural community in Alagoas.

Materials and Methods

Study Area and Population

Alagoas is the second smallest state of Brazil, situated in the northeast of the country, with a population of about 3 million. The economy of the state is based on agriculture, mainly sugar cane and coconut plantations, as well as fishing. The climate of Alagoas is tropical with intense rainfalls from April to August. During this period, 70% of the annual precipitation of 2,000 mm occurs. The average daily temperature is 26.6℃ in the dry season and 25.1℃ in the rainy season.

The study was carried out in Feliz Deserto, a typical village situated approximately 120 km south of Maceió, the capital of Alagoas. It is located at the coast and has a total population of 3,850 inhabitants.

The study area was defined as those districts of Feliz Deserto, where tungiasis commonly had occurred in the past (according to local health personnel, community leaders, and our own observations). A cohort of 1,250 individuals, about one third of the total population of the municipality, was involved in the study. Living conditions in these districts were more precarious than in the other areas of town.

Study Design

Two cross-sectional surveys were done: all households in the study area were visited in June/July 2003 (rainy season), and a second time in October/November 2003 (dry season). To be included in the study, individuals were required to spend at least four nights per week in a household in the study area. Anyone refusing to take part in the study was excluded. Individuals moving into the study area between the surveys were registered and took part in the second survey.

All household members were examined clinically for the presence of tungiasis. Numbers of lesions and associated pathology were documented on standardized questionnaires. Additionally, household members were interviewed to obtain socio-demographic and clinical data. If a household member was absent, the household was revisited three times. For some household members an appointment was made at work or at home. Each survey was carried out within a period of two months.

To reduce inter-observer bias, all clinical examinations were carried out by one investigator (A.J.).

Clinical Examination and Questionnaires

For the clinical examination, the head of the household was asked for a room with good light and in which privacy was guaranteed. In this room, the body was thoroughly examined for the presence of tungiasis.

Tungiasis was diagnosed if at least one embedded sand flea was present. Lesions were classified according to the Fortaleza classification [20]: a penetrating flea (stage I); an itching, reddish-brown spot with a diameter of one to three mm indicating the complete penetration of the flea into the epidermis (stage II); and a circular lesion presenting as a white patch with a central black dot and a diameter of four to ten mm indicating the maturity of the egg-expelling flea (stage III) are defined as viable lesions. Lesions with a black crust surrounded by necrotic tissue (stage IV), as well as partially or totally removed fleas leaving a characteristic sore in the skin are defined as nonviable tungiasis, i.e. the parasite is dead.

Lesions were counted and the topographic localization was documented on a visual record sheet.

According to Muehlen et al. [21] and Ugbomoiko et al. [22], a mild infestation was defined as the presence of 1 to 5, a moderate infestation as the presence of 6 to 30, and heavy infestation as the presence of >30 lesions.

Symptoms and signs associated with acute or chronic tungiasis were noted as previously defined [23]. Superinfection was diagnosed when pustules or suppuration were present. Socio-demographic data were assessed using pre-tested structured questionnaires.

Data Entry and Statistical Analysis

Data were entered twice into a database using the Epi Info software package (version 6.04d; Centers for Disease Control and Prevention, Atlanta, USA) and checked for entry errors. Then, data were transferred to Stata® software package (version 9.0; Stata Corporation, College Station, USA) for analysis. The Fisher's exact test was applied to determine the significance of differences of relative frequencies. As data were not normally distributed, the median and the interquartile ranges were used to indicate the average and the dispersion of data. Significance of differences between quantitative measurements was calculated by the Mann-Whitney test.

Ethical Aspects

Ethical clearance was obtained from the Ethical Committee of the School of Health Sciences of Alagoas (Escola de Ciências Médicas de Alagoas), the responsible ethical body in Alagoas State, as well as from an ad hoc ethical committee of the health department of the municipality of Feliz Deserto. Prior to the study, meetings were held with politicians, community leaders, and health care workers to explain the objectives of the study. All participants were informed about the study and gave their written consent. In the case of minors, the legal guardians were asked for written consent. Superinfected lesions were treated with topical antibiotic ointment or oral Roxithromycin (Floxid®, two doses of 150 mg over 5 days, Solvay Farma, São Paulo, Brazil). If other skin diseases were diagnosed during the surveys, the patients were referred to the primary health care centre of Feliz Deserto, where treatment is available free of charge.

Results

In the first survey in the rainy season (June/July 2003), 1,015 individuals of the target population of 1,146 (88.6%) were encountered and examined. During the second survey in the dry season (October/November 2003), 990 of 1,169 (91.1%) individuals were examined.

The overall prevalence of tungiasis was 21.6% in the rainy and 29.5% in the dry season ($p<0.0001$; Table 1). In both surveys, prevalence was highest in the children and decreased abruptly in the age groups ≥ 15 years. In the dry season, prevalence reached up to 40% in children between five and 14 years of age. Prevalence was already very high in children <5 years (Table 1).

The increasing prevalence in the dry season was most obvious in the adult age groups (20 to 39 and 40 to 59 years). In these age groups the prevalence of tungiasis doubled, as compared to the rainy season (Table 1). In contrast, in children <15 years, who were in general more affected, the relative increase in the dry season was less pronounced.

In both surveys, the overall prevalence of tungiasis was higher in males (rainy season: 23.5%; dry season: 32.2%), as compared to females (20.0% and 27.2%), but the difference was not statistically significant (p=0.19 and p=0.09, respectively).

Figure 1 depicts the prevalence by age groups and sex for the rainy and the dry season. Age-specific prevalence patterns are similar in both surveys, with higher prevalence in the dry season in almost all age groups. The highest prevalence was found in boys (5 to 14 years) in the dry season (48.3%; 95% confidence interval: 40.6-56.0; Figure 1). The age-specific prevalence followed an S-shaped curve. This pattern was most prominent when prevalence was highest, i.e. in the dry season.

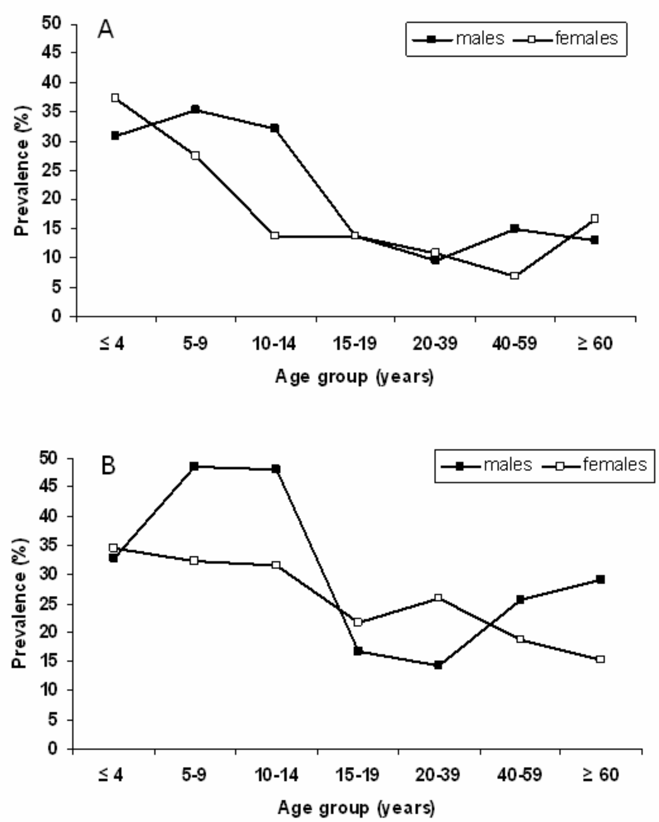

Figure 1. Prevalence of tungiasis, stratified by age group and sex. A. rainy season. B. dry season.

In total, 656 lesions were counted in 219 individuals during the rainy season, and 1,067 lesions on 292 individuals during the dry season. The median number of lesions was two in both surveys, with no statistical difference (p=0.5). The mean number of lesions in the rainy season was 2.99, and in the dry season 3.65 per case. A maximum of 42 lesions per individual was counted in the rainy season, and of 45 lesions in the dry season.

Table 1. Prevalence of tungiasis in the study area, stratified by age group and season.

Age group (y)	Rainy season (June/July) n positive	% (95% CI)	Dry season (October/November) n positive	% (95% CI)	p value
0 – 4	53/154	34.4 (26.9–42.5)	51/151	33.8 (26.3–41.9)	1.0
5 – 9	61/194	31.4 (25.0–38.5)	78/193	40.4 (33.4–47.7)	0.07
10 – 14	47/154	30.5 (23.4–38.4)	58/145	40.0 (31.9–48.5)	0.09
15 – 19	13/95	13.7 (7.5–22.3)	17/88	19.3 (11.7–29.1)	0.3
20 – 39	26/252	10.3 (6.9–14.8)	51/241	21.2 (16.2–26.9)	0.001
40 – 59	12/119	10.1 (5.3–16.9)	26/122	21.3 (14.4–29.6)	0.02
≥ 60	7/47	14.9 (6.2–28.3)	11/50	22.0 (11.5–36.0)	0.4
Total	219/1015	21.6 (19.0–24.1)	292/990	29.5 (26.6–32.3)	<0.0001

Table 2. Topographic distribution of embedded sand fleas.

Topographic site	Rainy season (n = 219) Number of lesions (%)	% of individuals with tungiasis[†]	Dry season (n = 292) Number of lesions (%)	% of individuals with tungiasis[†]
Feet:				
Toes without periungual sites	221 (33.7)	51.1	390 (36.6)	50.7
Toes periungual	302 (46.0)	69.4	539 (50.5)	70.6
Soles/plantar	93 (14.2)	18.3	96 (9.0)	14.4
Heels	15 (2.3)	3.7	13 (1.2)	3.8
Other	1 (0.2)	0.5	2 (0.2)	0.7
Ectopic sites:				
Hands	22 (3.4)	6.9	25 (2.3)	5.1
Other	2 (0.3)	0.9	2 (0.2)	0.7
All sites	656 (100)	100	1067 (100)	100

[†] Lesions occurred on more than one site.

In the rainy season 190/219 (86.8%), and in the dry season 246/292 (84.2%) individuals with tungiasis had a mild infestation (1–5 embedded sand fleas). Only one (0.5%) individual in the rainy and five (1.7%) in the dry season were heavily infested (>30 lesions). Interestingly, the only individual in the rainy season with heavy infestation, representing about 1/200 of the cases with tungiasis, had 42 lesions and thus was responsible for 6.4% of all lesions in the study population. In the dry season, the five most heavily infested individuals, despite representing less than 2% of the infested population, harboured 17.3% of the total parasite load in the community.

Table 3. Tungiasis-associated symptoms and signs.

Clinical feature	Rainy season (n = 219) n	%	Dry season (n = 292) N	%
Desquamation of skin	126	57.5	130	44.5
Hyperkeratosis	113	51.6	101	34.6
Nail deformation	70	32.0	68	23.3
Sleep disturbance due to itching	64	29.2	61	20.9
Secondary infection	34	15.5	40	13.7
Oedema	27	12.3	20	6.9
Erythema	27	12.3	22	7.5
Pain upon pressure	27	12.3	34	11.6
Fissure	23	10.5	27	9.3
Constant pain	20	9.1	20	6.9
Ulcer	14	6.4	9	3.1
Loss of toe-nail(s)	12	5.5	7	2.4
Inguinal lymphadenopathy	8	3.7	24	8.2
Difficulty walking	3	1.4	2	0.7
Deformation of digits	2	0.9	5	1.7

Most lesions occurred on the feet (rainy season: 96.3%; dry season: 97.5%). However, a considerable number of individuals presented with lesions on the hands (Table 2). About 70% of infested individuals had periungual lesions, and 50% on the toes on topographic areas other than periungual.

The most common clinical signs in individuals with tungiasis were desquamation of skin and hyperkeratosis. Deformation of nails was also very common (Table 3). About 15% of cases showed signs of superinfection. Severe pathology, such as deep fissures, and difficulty walking was also common. Interestingly, 67% of the individuals stated that they felt the flea penetrating into the skin.

Discussion

Our data show that tungiasis is prevalent in the rural community under study, and that prevalence varies according to the season. Children were identified as a high-risk group. The prevalence of tungiasis encountered in Feliz Deserto (22% in the rainy season, 30% in the dry season) was impressively high, although considerably lower as compared to other studies in impoverished communities. For example, in population-based studies in rural Brazil and Nigeria, prevalences of 51% and 45% were found [6,9]. In an urban slum in Brazil, prevalence in the peak dry season was as high as 54% [24]. Njeumi et al. (2002) reported about 50% of school children to be infested in different communities in Cameroon [14].

The relatively low prevalence found in our study may have different reasons. Housing was much better than in the other impoverished communities studied: in Feliz Deserto all houses had access to piped water, and most had cemented floors. A sandy floor inside the house is known as a major risk factor for the presence of tungiasis and severity of infestation [21,22]. In Feliz Deserto, pigs were rare and not allowed to roam freely within the community - free-roaming pigs were common in a Nigerian community with a high prevalence and contributed to an important degree to transmission [22]. Since waste collection was performed daily, there was little waste littering the inhabited area, reducing the number of rats serving as an animal reservoir [25].

In Feliz Deserto the most severely affected age groups were children < 15 years of age, irrespective of the season. This seems to be a characteristic finding pertaining to many endemic areas [4-6,8-10,26]. Tungiasis has to be considered mainly a disease of the childhood.

The S-shaped age-specific distribution with peak prevalences in children and the elderly was less pronounced in Feliz Deserto than in other endemic communities [6,9,15]. Particularly, the increase of prevalence in the elderly was less accentuated. However, when age-related prevalences were stratified into males and females a peculiar pattern emerged: whereas prevalence in females decreased constantly with increasing age, in males the S-shaped curve was very clear.

The increased prevalence in the elderly male indicates that acquired immunity against *T. penetrans* is unlikely to develop. Re-increasing prevalences in elder male age groups are likely due to differences in exposure between men and women. In Feliz Deserto, adult men frequently work outside the endemic area during the day and commonly use closed shoes during work. In contrast, elderly males are usually retired, stay the whole day in the community, walk barefooted, and put their bare feet on the ground when sitting and chatting. Alternatively, they may care less about penetrated fleas, while elder women tend to take out sand fleas as soon as they perceive a penetrated flea.

In our study, the prevalence of tungiasis was slightly higher in the male sex in both surveys. Apparently, the pattern of occurrence of tungiasis in males and females differs from community to community. For example, Carvalho et al. (2003) observed significantly more females than males affected in a resource-poor community in South Brazil [5]. No difference in the prevalence between males and females was found in a rural community in Lagos State in Nigeria, and in a fishing community in Ceará State, Brazil [6,9]. In contrast, authors from Brazil, Trinidad, and Nigeria found that males were predominantly affected [4,8,10,26]. In a longitudinal study in an urban slum, tungiasis was consistently more common in the male sex during a study period of 12 months [24].

The parasite load and the maximum number of lesions found in Feliz Deserto were rather low (mean < 4 in both surveys). In a fishing community in Brazil, the mean parasite load was 8.9 [9], in a rural community in Nigeria 12.3 [6], in an urban slum in Ceará State 7.8 [10], and in several communities in Trinidad 8.0 [4]. In the latter two studies, prevalence was comparable to Feliz Deserto with 34% and 21%, respectively. Thus the hypothesis formulated previously that prevalence and intensity of infestation are positively related did not hold true for Feliz Deserto [9]. As it is known that a high exposure is correlated with a high parasite load [27], we assume that in Feliz Deserto exposure and transmission rates are low, as compared to the other endemic communities studied.

Parasite load was disproportionately distributed with only few individuals harbouring a high number of lesions, whereas the majority had only one or two embedded sand fleas. In the dry season, only five individuals were responsible for 17% of the total parasite load in the community. Similar patterns have been observed previously [6, 9,11,15]. For example, in a fishing community, 8% of infested individuals accounted for 55% of the total parasite load in the community [9]. The rather few severely infested individuals may contribute disproportionately to the excretion of eggs by embedded sand fleas and therefore are responsible for maintaining transmission dynamics at a high level. Thus, identifying the most severely infested individuals in a community and focusing control on their households could be an efficient way to reduce transmission rates, as well as the degree of morbidity in an endemic area.

In our study, the topographic distribution of sand flea lesions was similar to other previously published studies [6,9]: about 70% of individuals presented with lesions on periungual sites of the feet. Less than 5% of lesions occurred on ectopic sites, i.e. other topographic areas than the feet. The topographic distribution did not vary considerably between rainy and dry season, indicating that sand fleas prefer certain predilection sites, irrespective of environmental conditions. The number of ectopic lesions was lower, as compared to communities with higher prevalences in the general population. In these populations, lesions occurred on sites other than the feet in about 10% of cases [6,9]. As ectopic tungiasis is more common in individuals with a high parasite load [28], and because on the population level intensity of infestation is positively related to the prevalence, the occurrence of ectopic lesions could be used as a predictor for the prevalence of tungiasis in a community. As ectopic lesions, e.g. on the hands, are easily diagnosed, the presence of ectopic sand fleas may be useful as a rapid assessment method.

In the study area, nail deformations were frequent. Nail deformation is the consequence of repeated infestation with or without secondary infection. Nail deformation occurred in about 27% of cases. Severe sequels, such as difficulty walking and deformation of digits due to severe infestation existed, but were less common. This confirms the notion that tungiasis cannot be regarded as nuisance, but as an important health problem [6,16,29]. In Haiti, deaths were reported as a result of severe infestations, probably due to septicaemia and tetanus [7]. In rural Nigeria, difficulty walking occurred in 32%, deformation of toe-nails in 27%, and loss of toe-nails in 16% of the patients [6]. Between 20% and 29% of individuals infested with tungiasis stated that they woke up at night due to itching at the site of penetration. Itching has been repeatedly described as a predominant symptom of tungiasis [6,29,30]. However, other parasitic skin diseases, such as cutaneous larva migrans and scabies, are endemic in the study area [31,32], and polyparasitism may have occurred. Thus it is possible that these diseases also caused sleep disturbances in some individuals with tungiasis.

Our data show that the prevalence of tungiasis was significantly higher in the dry than in the rainy season. The difference was less pronounced than the seasonal variation observed in the only longitudinal study published so far [24]. Similar to our findings, in the latter study the highest prevalence was found in an urban slum in Brazil in the middle of the dry season. Prevalence differed by more than a factor three throughout the year (54% vs. 17%), and was clearly correlated to precipitation patterns [24]. Interestingly, in our study, in adults aged 20 to 59 years prevalence was twice as high in the dry season, as compared to the rainy season. This indicates that attack rates actually increased in the dry season. Seasonal variation of attack rates and incidence probably is caused by biological dynamics of the sand flea population, reflecting changes of environmental variables. During the rainy season, high soil humidity may impair the development of free-living stages of sand fleas, and heavy precipitations will wash away eggs, larvae, pupae, and adult stages from the area where they developed [24].

The varying incidence of *T. penetrans* during the year is a good explanation for the highly diverging prevalences observed in previous population-based studies. In fact, studies were performed in different seasons, and sometimes over a prolonged period of the year; and often period rather than point prevalences were assessed [4-6,8,9,11-15,33]. In the present study, each survey lasted about two months. Thus, we cannot exclude that the attack rate may have changed over time and that prevalences calculated are not true point prevalences. However, even if this error occurred, it should be irrelevant, as climate conditions did not change during each survey.

In the present study, we reduced participation bias by organizing meetings with the community members before starting the study. During field work, all individuals were invited to present at the health care centre, if they were not encountered at home, and households were visited three times. Households were visited also on weekends to increase the participation of working males. However, non-participation of these groups was still an issue. We excluded inter-observer bias, as only one investigator realized all clinical examinations in both surveys. Intra-individual bias was reduced by doing clinical pilot studies before the survey, and by rigorously defining the clinical criteria of tungiasis.

In conclusion, our study shows that tungiasis is common in a typical resource-poor community in Alagoas State, and that prevalence varies according to the season. The most vulnerable group was children. Tungiasis was associated with considerable morbidity. Few individuals contributed disproportionately to the total parasite load of the community and could be a good target for interventions.

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