

## ECTOPIC LOCALIZATION OF TUNGIASIS

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**Abstract.** Tungiasis is caused by the penetration of the female sand flea *Tunga penetrans* into the epidermis. It is generally assumed that lesions are confined to the feet. To determine to what degree tungiasis occurs at other topographic sites, 1,184 inhabitants of a poor neighborhood in northeastern Brazil were examined; 33.6% were found to have tungiasis (95% confidence interval = 30.9–36.4%). Six percent presented lesions at locations other than the feet, with the hands being the most common ectopic site (5.5%). Other sites were the elbows, thighs, and gluteal region. Ectopic tungiasis was significantly associated with the total number of lesions ( $P < 0.001$ ) and an age less than 15 years old ( $P = 0.02$ ). In 86 patients actively recruited with lesions on their feet, ectopic localizations were observed in 25.6%. Since untreated sand flea lesions are prone to become superinfected, clinicians should be aware of not missing any ectopic localization of tungiasis.

### INTRODUCTION

Tungiasis is an ectoparasitic disease caused by the penetration of the female sand flea *Tunga penetrans* into the epidermis of its host. Beside humans, various domestic and sylvatic animals are affected.<sup>1</sup> The sand flea, also called the jigger, originally occurred on the South American continent and the Caribbean Islands, but was inadvertently introduced into sub-Saharan Africa in the late 19th century.<sup>2,3</sup> It is endemic in many countries in Latin America, the Caribbean and sub-Saharan Africa.

Since the 1970's, probably due to growing urbanization and improved housing, the occurrence of the sand flea seems to have decreased. However, tungiasis remains an important health problem of people living in severe poverty. In Brazil, prevalence rates reach up to 40% in the miserable squatter camps at the outskirts of the big cities, as well as in the underdeveloped rural hinterland.<sup>1</sup>

In endemic areas, particularly in children, tungiasis is associated with severe morbidity.<sup>1</sup> Without appropriate treatment, secondary infections with pathogenic bacteria are extremely common (Heukelbach J and others, unpublished data). In severe cases, sepsis, lymphatic edema, and auto-amputation of digits can occur.<sup>4–8</sup> Tungiasis also has been associated with tetanus in non-vaccinated individuals.<sup>9,10</sup>

Since *T. penetrans* is unable to jump high, it is generally assumed that lesions are confined to the feet. This, in turn, causes physicians to restrict their attention to the lower extremities of patients with tungiasis.

To determine whether tungiasis may occur at other sites than the feet, we examined a representative sample of a population living in a highly endemic area, as well as heavily infested patients actively recruited at the Primary Health Care Center (PHCC) in a Brazilian community.

### MATERIALS AND METHODS

**Study area.** The study was performed in the *favela* of Serviluz, a typical shantytown at the outskirts of Fortaleza, the capital of Ceará State in northeastern Brazil. This *favela* is close to the beach, where poor immigrants from the arid interior have started to invade the dunes and to construct houses since the 1950s. The area has a total population of approximately 15,000 inhabitants. Ninety-seven percent of the households are connected to the electric

power supply, and 70% have access to piped water. Sixty percent of the population have a monthly family income of less than two minimum wages (one minimum wage = US\$ 60).<sup>11</sup> Adult illiteracy is 30%, and crimes of all sorts are common.

The *favela* is divided into five areas, for each of which detailed demographic data are available from a recently performed census. Three of the five areas are considered by the personnel of the PHCC to be heavily affected. Of these, the area of *Morro de Sandras* was randomly selected.

**Study population and examinations performed.** All 327 families in *Morro de Sandras* were visited by one of the investigators (TW). In each household, informed oral consent was obtained, and each member of the family was thoroughly examined. In case of absent family members, the household was visited a second time. A total of 1,184 (80.7%) of 1,467 inhabitants was included in the study. This study has been approved by the Ethical Committee of the Federal University of Ceará, Fortaleza, Brazil. The committee responsible for medical investigations of the community (*associações dos moradores*) agreed also to perform the study. Additionally, permission was obtained from the Health Secretary of Fortaleza Municipality and from the Ministry of Health of Ceará State, Brazil. The physicians and the medical staff of the PHCC were informed and agreed to co-operate. Prior to the study, meetings with community health workers and community leaders were held in which the objectives were explained. Informed oral consent was obtained from each patient after explaining the objectives of the study. In the case of minors, the caretakers were asked for consent.

The entire body of each participant was systematically examined. The following findings were considered to be diagnostic for tungiasis: a red-brown itching spot with a diameter of 1–2 mm (early stage), lesions presenting as a white patch with a diameter of 3–7 mm with a central black dot (mature stage), a dead flea with surrounding necrosis (late stage), as well as lesions altered through manipulation by the patient (e.g., partly or totally eliminated fleas leaving a characteristic sore in the skin and suppurative lesions caused by the use of non-sterile perforating instruments such as needles and thorns). Localization, stage, and number of lesions were documented. The presence of scabies, head lice, and cutaneous larva migrans was also recorded. In addition, 86 patients with severe tungiasis were actively recruited by the commu-

nity health workers of the PHCC. These patients were examined in an identical manner.

**Data analysis.** Data were entered into a database using Epi Info software package (version 6.04d; Centers for Disease Control and Prevention, Atlanta, GA) and checked for validity. The random number generated from the Epi Info software was used to select one of the three hyperendemic areas. Risk ratios (RRs) to identify risk factors for the presence of ectopic tungiasis and 95% confidence intervals (CIs) of point prevalence rates were calculated using the respective Epi Info modules. Chi-square and Wilcoxon (Mann-Whitney) tests were used where appropriate.

## RESULTS

Three hundred ninety-eight individuals (33.6%) from *Morro de Sandras* had tungiasis (95% CI = 30.9–36.4%); 25% were children 5–9 years old (prevalence rate = 56.5%, 95% CI = 48.7–64.2%). Significantly more males than females showed the presence of sand fleas (43.7%; 95% CI = 39.4–48.1% versus 25.7%; 95% CI = 22.4–29.2%,  $P < 0.001$ ).

Twenty-three patients (5.8%) presented with tungiasis at topographic sites other than the feet. Of 3,120 lesions recorded in 1,184 individuals, 101 (3.2%) were at ectopic sites. Twenty-two patients showed lesions on the hands (5.5%), three on the elbows (0.8%), and one on the thighs (0.3%; Table 1). Three patients had ectopic tungiasis without lesions on the feet.

Risk factors for ectopic localization were the presence of five or more lesions (RR = 4.8,  $P < 0.001$ ) and an age less than 15 years old (RR = 4.8,  $P = 0.02$ ). However, 2.3% of the patients with less than five lesions also had ectopic tungiasis. In individuals more than 15 years old, only two cases of ectopic tungiasis were observed. Sex was not correlated with the presence of ectopic lesions.

In the 86 patients actively recruited by community health workers, ectopic localizations were even more common. Twenty-one had *T. penetrans* lesions on the hands (24.4%) and one patient had lesions in the gluteal region (1.2%). The mean number of lesions was 28 compared with 2.6 in the community-based study ( $P < 0.001$ ).

## DISCUSSION

Ectopic localizations of tungiasis have been previously reported as extremely rare. Case reports from Brazil and other countries suggest that ectopic localizations occur in patients

with severe infestations. Bezerra described a homeless beggar with severe tungiasis who had multiple lesions on the legs and hands.<sup>12</sup> In southern Brazil, ectopic localizations have been observed on the hands, legs, trunk, and face.<sup>13</sup> Connor reported that even infestation of the eyelids could occur.<sup>14</sup> In Trinidad, a point prevalence rate of lesions on the hands of 9.8% has been reported in patients with tungiasis.<sup>5</sup> We have observed lesions on the testes, penis, inguinal area, neck, trunk, and abdomen in patients from other areas of the *favela* (Heukelbach J and others, unpublished data). Interestingly, multiple pubic and inguinal lesions have been reported in a man who had been sitting on a sack containing infested sisal hemp from Mexico; the embedded fleas were confined to these sites.<sup>15</sup> Travelers in tropical Africa have shown lesions on the back, under the breast and on the wrist after using bed sheets that had hung outside touching the ground.<sup>16</sup>

The assumption that tungiasis is a disease of the feet is corroborated by its popular name. In Brazil, the sand flea is called “*bicho de pé*,” literally, “beast of the feet.” Clearly, both our experience and that of others has shown that infestation can occur in virtually any site of the body.<sup>5,12–16</sup>

Although it has been reported that *T. penetrans* cannot jump high and consequently chooses the feet as the preferred site for penetration into the epidermis,<sup>16–19</sup> other factors hitherto unknown may lead to aggregation of lesions in the toes or heels. We have frequently observed fleas running across the skin with a mean velocity of approximately 10 mm/sec (Heukelbach J and others, unpublished data). However, the mechanism that causes fleas to migrate from the soil to distant parts of the body remains enigmatic.

The presence of ectopic tungiasis is associated with the total number of lesions and a young age. Ectopic lesions occurred almost exclusively in children less than 15 years of age. This can be explained by the fact that the children of the *favela* usually play in unpaved streets and backyards, which consist mainly of sand, and thus not only have high exposure rates but also bring body parts other than the feet in contact with contaminated soil. In fact, children of all age groups are more frequently infested with *T. penetrans* than adults and also have more lesions (Heukelbach J and others, unpublished data).

Patients actively recruited by the community health workers had ectopic lesions four to five times more frequently than individuals in the community study. This is not surprising, since the health workers selected heavily infested patients and referred them to the PHCC. Whereas the average number of lesions was 2.6 in the community-based study, it was 28 in the actively recruited patients.

In endemic areas, the diagnosis of tungiasis is exclusively made through macroscopic inspection of the lesion. Although physicians sometimes have difficulties in diagnosing tungiasis in travelers returning from the tropics and request histopathologic examination of a biopsy sample,<sup>20–28</sup> the etiology of the lesion, particularly its dynamic changes in size and macroscopic appearance, is so characteristic that the diagnostic findings used in this study are unlikely to be inaccurate and are believed to be virtually pathognomonic for tungiasis. However, it cannot be excluded that other lesions have been overlooked or misdiagnosed.

Although there are dozens of case reports of tungiasis in travelers returning from tropical countries,<sup>6,8,17,20–28</sup> ectopic localizations have been reported in only two cases.<sup>16,19</sup> It is

TABLE 1

Topographic localization of tungiasis in the community study group (n = 1,184)

Topographic site	Number positive*	% of individuals with tungiasis
Feet		
Periungueal	341	85.7
Other sites (heels, lateral rim, plantar)	151	37.9
Hands	22	5.5
Elbows	3	0.8
Thighs	1	0.3
Any ectopic site	23	5.8
Tungiasis at any site	398	100

\* Multiple lesions of more than one site were common.

unlikely that a traveler with tungiasis will have overlooked a lesion on his or her body. In addition, few lesions are usually observed in tourists. This suggests an association between heavy infestation and fleas located at ectopic sites. Since untreated lesions are prone to bacterial superinfection, physicians working in areas endemic for tungiasis should be aware of examining patients from the feet to the head so that fleas embedded at ectopic sites are not overlooked.

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