doi: 10.5216/rpt.v51i1.70027

REVIEW

PREVALENCE OF TUNGIASIS IN HUMANS IN BRAZIL AND IN ITS FEDERATIVE UNITS: A SYSTEMATIC REVIEW

Laura Silva Paranhos[1], Pedro Alves Soares Vaz de Castro[2], Giovanna Rotondo de Araújo[3,4], Fernanda do Carmo Magalhães[4] and Juliana Maria Trindade Bezerra[4,5,6]

ABSTRACT

Tungiasis is a neglected parasitic disease caused by penetration of female *Tunga penetrans* under the skin, causing important health outcomes in humans. Therefore, the aim of this study was to describe the prevalence of tungiasis in Brazil and in its federative units. In November 2019, an investigation was carried out to find studies published from 1980 onwards in MEDLINE, LILACS, Cochrane, CINAHL, Scopus, Web of Science and Embase databases, and in the gray literature, using descriptors related to the prevalence of tungiasis caused by *T. penetrans* in Brazil. Of the 542 studies found, only 16 published between 2002 and 2010 met the eligibility criteria to be included in this systematic review. Of the 16 selected publications, 14 addressed the prevalence of tungiasis in communities in the Northeast region of the country, one in the South and one in the Southeast. The general prevalence of the parasitosis in the studies ranged from 1.6% to 54.8%, predominantly in the five to nine age group. Eight studies considered the prevalence by gender, ranging from 2.2% to 62.2% for females and 1.1% to 62.5% for males. This systematic review presents an unprecedented survey of the prevalence of tungiasis, a parasitic disease whose dissemination is facilitated by several factors, occurring mainly in low-income communities. Considering the regionalization of the findings, the scarcity of publications, as well as disease neglect, more studies are required.

KEY WORDS: *Tunga penetrans*; occurrence; epidemiology; PRISMA; human.

1. Universidade Federal de Minas Gerais, Curso de Bacharelado em Enfermagem, Belo Horizonte, Minas Gerais, Brazil.
2. Universidade Federal de Minas Gerais, Curso de Bacharelado em Medicina, Belo Horizonte, Minas Gerais, Brazil.
3. Universidade Federal de Minas Gerais, Laboratório de Epidemiologia e Controle de Doenças Infecciosas e Parasitárias, Departamento de Parasitologia, Instituto de Ciências Biológicas. Belo Horizonte, Minas Gerais, Brasil.
4. Universidade Federal de Minas Gerais, Programa de Pós-Graduação em Parasitologia, Departamento de Parasitologia, Instituto de Ciências Biológicas, Belo Horizonte, Minas Gerais, Brazil.
5. Universidade Estadual do Maranhão, Curso de Licenciatura em Ciências Biológicas, Centro de Estudos Superiores de Lago da Pedra, Lago da Pedra, Maranhão, Brazil.
6. Universidade Estadual do Maranhão, Programa de Pós-Graduação em Ciência Animal, São Luís, Maranhão, Brazil.

Laura Silva Paranhos: https://orcid.org/0000-0002-5941-7949; Pedro Alves Soares Vaz de Castro: https://orcid.org/0000-0002-1183-7610; Giovanna Rotondo de Araújo: https://orcid.org/0000-0002-7513-5087; Fernanda do Carmo Magalhães: http://orcid.org/0000-0001-7928-8294; Juliana Maria Trindade Bezerra: https://orcid.org/0000-0001-9840-6217

Corresponding author: Juliana Maria Trindade Bezerra. Centro de Estudos Superiores de Lago da Pedra, Universidade Estadual do Maranhão, Avenida Roseana Sarney, s/n, Vila Rocha, CEP 65.715-000 Lago da Pedra, Maranhão, Brasil. E-mail: julianamtbezerra@outlook.com

Received for publication: 24/8/2021. Reviewed: 8/2/2022. Accepted: 23/3/2022.

INTRODUCTION

Tungiasis is a neglected parasitic disease caused by penetration of female *Tunga penetrans* (Linnaeus, 1758) (Siphonaptera; Tungidae) (sand flea or jigger flea) under the skin. This flea is often reported as the only zoonotic species in Brazil (Linardi et al., 2013). However, *Tunga trimamillata,* first discovered in Ecuador and Peru, has also been described as cause of infections in humans and domestic animals in Brazil (Pampiglione et al., 2009; Linardi et al., 2013).

The flea *T. penetrans* is the smallest known flea species, only a millimeter in size, and although males and females feed on blood (Witt et al., 2004), only females permanently penetrate the skin of their hosts and suffer significant hypertrophy, expelling hundreds of eggs over a period of two to three weeks (Eisele et al., 2003). The periungeal region of the toes is the preferred location for the flea, although infestation can also occur in the hands, elbows, and genital and anal regions (Feldmeier et al., 2003).

Tungiasis is usually a self-limiting infestation; however, some complications are common in endemic areas (Feldmeier et al., 2003; Heukelbach et al., 2001). Sequelae include severe pain, inflammation, cracking, loss of toenails, as well as finger deformity (Feldmeier et al., 2003). The skin wound caused by the ectoparasite can also serve as an entry point for pathogenic microorganisms, such as *Staphylococcus aureus, Streptococcus* spp. and *Clostridium* spp. (Feldmeier et al., 2002).

Some climatic, socioeconomic, and cultural factors directly influence infestation rate (Mazigo et al., 2012), which is more prevalent in children, particularly in five to ten-year-olds (Mazigo et al., 2012; Heukelbach et al., 2007). Associated with poverty, it occurs in many communities characterized by scarce financial resources, low educational level (Heukelbach et al., 2001; Heukelbach et al., 2002; Muehlen et al., 2006), poor personal hygiene, lack of sanitation in homes and the residential environment, as well as the non-use of shoes (Muehlen et al., 2006; Nair et al., 2016; Ugbomoiko et al., 2007). Furthermore, the presence of reservoirs for the parasite in the peridomestic environment, such as pigs, cats, dogs, and rats, is also an important risk factor (Franck et al., 2003; Heukelbach, 2005; Heukelbach et al., 2005b).

Tungiasis is common in low-income communities in tropical and subtropical parts of the world, such as the Caribbean, South America, and Africa (Heukelbach, 2005; Heukelbach et al., 2005c). In Brazil, it is popularly known as sand flea, foot bug or pig bug, with prevalence rates from 16% to 55%. It has been documented in several communities from the Amazon region to the South, being particularly present in precarious urban settlements, rural areas, and fishing communities (Matias, 1989; Muehlen et al., 2003; Carvalho et al., 2003; Heukelbach et al., 2004a; Ariza et al., 2007; Damazio & Silva, 2009). Despite being an anthropozoonosis, it is

neglected by authorities in spite of being a serious health issue, proving to be a little-known disease. This systematic review addresses the prevalence and epidemiological aspects related to tungiasis in humans in Brazil and its federative units.

METHODS

Study area

Brazil, officially the Federative Republic of Brazil, is the largest country in South America and Latin America, being the fifth largest in the world in territorial area (comprising 47.3% of the South American territory) and sixth in population (with more than 211 million inhabitants). Bordered by the Atlantic Ocean to the east, Brazil has a coastline of 7,491 km. The country borders all other South American countries, except Chile and Ecuador. In the north by Venezuela, Guyana, Suriname, and the French overseas department of French Guiana; Northwest by Colombia; in the West by Bolivia and Peru; in the Southwest by Argentina and Paraguay and in the South by Uruguay.

The country consists of 26 States and a Federal District, with 5,570 municipalities spread over five regions: North (comprising seven States: Acre, Amapá, Amazonas, Pará, Roraima, Rondônia, and Tocantins), Northeast (comprising nine States: Alagoas, Bahia, Ceará, Maranhão, Paraíba, Pernambuco, Piauí, Rio Grande do Norte, and Sergipe), Midwest (comprising three States: Goiás, Mato Grosso, and Mato Grosso do Sul; in addition to the Federal District), Southeast (comprising four States: Espírito Santo, Minas Gerais, Rio de Janeiro, and São Paulo) and South (comprising three States: Paraná, Rio Grande do Sul, and Santa Catarina) (IBGE, 2021).

Study question

The guiding question for the study was the following: "What is the prevalence of tungiasis caused by *T. penetrans* in Brazil and in its 27 federative units?" The study question was defined considering the PICOS anagram (Methley et al., 2014), where P represents the Population (people affected by tungiasis caused by *T. penetrans* in Brazil and in the 27 federative units); I represents the Intervention (or exposure) (not applicable); C is related to Comparison (not applicable); Outcome represents the prevalence of tungiasis in Brazil and in its 27 federative units; and S represents the Study Design (observational epidemiological studies).

Study design and protocol registration

The present study is a systematic literature review. This study was registered in the International Prospective Register of Systematic Reviews (PROSPERO) under registration number CRD42020147957 (PROSPERO, 2020). This systematic review followed the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Page et al., 2021).

Eligibility criteria

Observational epidemiological studies performed in Brazil, published from 1980, that described the prevalence of tungiasis in Brazil and in its 27 federative units, available in English, Spanish or Portuguese, were considered eligible for this systematic review. There were no restrictions on the characterization of study participants. The searches were carried out on November 26, 2019, and the surveys were updated until June 22, 2021, to verify any possible new studies published on the researched topic. Studies not conducted in Brazil and/or in its 27 federative units, that did not correspond to the languages adopted in the search strategies, or that did not fit the study question were excluded.

Research sources and strategies

Investigations were performed in the electronic scientific databases MEDLINE [via US National Library of Medicine (PubMed)], LILACS (Latin American and Caribbean Literature on Health Sciences) via the regional portal of the Virtual Health Library (VHL), Cochrane, CINAHL, Scopus, Web of Science and Embase. Bibliographic reference lists of relevant studies were examined to identify eligible papers.

Searches were performed using descriptors listed in the *Descritores em Ciências da Saúde* (DeCS) and Medical Subject Headings (MeSH), in English, Spanish and Portuguese. Studies that contained the descriptors in the title, abstract and keywords were selected. The Boolean operators "AND" and "OR" were used, as well as quotation marks to facilitate the search for manuscripts. In each of the databases, word combinations were used, together or separately, in the three languages mentioned above: "Tungíase" (alternative terms: "*Tunga*", "*Tunga penetrans*", "Bicho-de-pé", "Bichos-de-pé", "Tungiasis"); "Prevalência" (alternative terms: "Prevalence", "*Prevalencia*"); "*Incidência*" (alternative terms: "Incidence", "*Incidencia*"); "*Epidemiologia*" (alternative terms: "Epidemiology", "*Epidemiología*"); and "*Fatores de Risco*" (alternative terms: "Risk Factors", *"Factores de Riesgo*"). After organizing strategies, the material was investigated separately by three researchers.

In order to avoid an inadequate or non-comprehensive selection, which would reduce the representativeness of the identified or included studies, the gray literature was also searched by Google Scholar access to find additional publications not detected in the selected electronic databases.

Selection of studies and data extraction

For study selection and data extraction, after removing duplicate records, three independent researchers selected the articles by title, abstract and full text, in separate and sequential steps, following the predefined inclusion and exclusion criteria. The three researchers independently selected the studies in the three stages and later met to verify agreement on inclusion and exclusion. There was no divergence in opinions among the three researchers.

After these steps, information was collected from the full texts (selected studies). The following data were extracted from the studies: authorship; year; location of the study by identification of the city and the federative unit; study design; database where the study was found; publication journal; study period, considering the first and last year of the research; study objectives; number of participants; gender, age group, income, place of residence, as well as other participant information; overall prevalence of tungiasis in the study; diagnosis of tungiasis; treatment of participants; statistical analysis used in the study; secondary illnesses due to tungiasis; limitations of the study; outcomes.

Assessment of the methodological quality of the included studies

Three criteria were considered, based on the Newcastle-Ottawa Scale (NOS) for the 16 cross-sectional studies to evaluate the methodological quality of the scientific studies included in this systematic review, (Wells et al., 2009): (i) selection (items evaluated: representativeness of the sample, sample size, non-respondents and determination of exposure), (ii) comparability (control for confounding factors); and (iii) outcome (evaluation of results and statistical tests adopted). Item (i) consisted of 5 questions, for which the evaluated studies could score from 0 to 5, with one point per question; item (ii) consisted of 2 questions, for which the evaluated studies could score from 0 to 2, with one point per question; and, item (iii) consisted of 3 questions, for which the evaluated studies could score from 0 to 3, with one point per question. The sum of the three items for each article, with their respective specific questions regarding each evaluation level (selection, comparability and outcome), therefore, ranged from zero to 10 points. In this systematic review, studies were considered of high quality when they obtained a value greater than or equal to five points (average, cross-sectional studies), according to the previously published NOS assessment definition (Luchini et al., 2017).

Data analysis

For table and graph organization, Microsoft Word and Excel 2013 packages were used (Redmond, Washington, United States of America) and for the preparation of the thematic distribution map of the studies included in the systematic review, for Brazil and its 27 federative units, QGIS 3.10 software (Norden, Germany) was used. The shapefile map was taken from the platform of the Regional and Urban Economy Center of the State of São Paulo (NEREUS, 2020).

RESULTS

Study Analysis

Initially, 541 studies were selected in the search within the seven electronic databases and one other in the gray literature (Google Scholar), totaling 542 studies. Two hundred and sixty-one studies were excluded due to duplication. 281 studies were selected according to title and abstract, of which 254 were excluded for not being related to the study question. The remaining 27 studies were read considering the full text and eleven were excluded for not answering the study question either. Thus, 16 studies were utilized in the present systematic review (Figure 1).

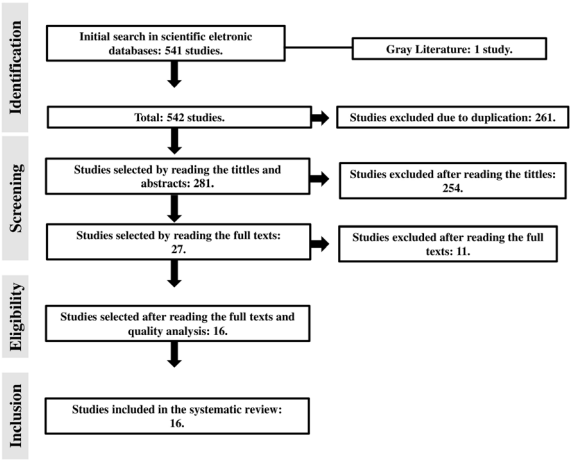

Figure 1. Flowchart of selection of scientific studies in the databases for the systematic review on the prevalence of tungiasis in Brazil and in its 27 federative units.

Characterization of the studies

Sixteen studies met the eligibility criteria and were selected for the present systematic review that covered three of the five Brazilian regions. The prevalence of tungiasis was addressed in communities in the States of the Northeast region in 14 studies (11 in the State of Ceará, one in Rio Grande do Norte, one in Alagoas, and one in the States of Ceará and Alagoas). The remaining two studies present data regarding communities in the States of Rio de Janeiro (Southeast region), and Santa Catarina (South region). All studies followed cross-sectional designs and were published between 2002 and 2010 (Figure 2 and Table 1).

Figure 2. Distribution of published scientific studies on factors associated with the prevalence of tungiasis, from 1980 to 2021, according to the Brazilian regions, where each study was carried out. N = Number of studies.

Prevalence and epidemiological aspects of tungiasis

The prevalence of tungiasis in the studies, considering the total number of participating individuals, ranged from 1.6% to 54.8% (Table 1). The prevalence by gender was described in eight of the 16 studies, ranging from 2.2% to 62.2% for females and from 1.1% to 62.5% for males. Age groups were heterogeneously characterized among the studies. However, the majority reported that the most prevalent infestation was in the 5-9 age group. The most common stratification and prevalence was 0-4 years (10.1% to 67.8%), 5-9 years (15.3% to 76.3%), 10-14 years (13.3% to 68.4%), 15-19 years (7.5% to 43.9%), 20-39 years (2.9% to 34.9%), 40-59 years (6.2% to 44.3%) and 60 years or older (9.7% to 57.9 %).

Table 1. General prevalence and seasonal variation in the prevalence of tungiasis determined in scientific studies on the parasitosis in Brazil.

Authors and year	Study location	Study design	Total number of participants (N)	Prevalence (95% CI)	Seasonal variation			
					Number of participants		Prevalence	
					Dry season	Rainy season	Dry season	Rainy season
Heukelbach et al. (2002)	Morro do Sandras, Fortaleza – State of Ceará	Cross-sectional population-based study	1,184	33.6% (30.9-36.4)	-	-	-	-
Wilcke et al. (2002)	Morro do Sandras, subarea Vicente Pinzón II, Fortaleza – State of Ceará	Cross-sectional study	1,185	33.6% (30.9-36.4)	-	-	-	-
Carvalho et al. (2003)	Bananeiras, Araruama – State of Rio de Janeiro	Cross-sectional study	241	54.8%	-	-	-	-

Heukelbach et al. (2003)	Morro do Sandras, subarea Vicente Pinzón II, Fortaleza – State of Ceará	Cross-sectional study	Community: 1,185 Primary Health Care Center: 288	Community: 33.6% (30.9-36.4) Primary Health Care Center: 19.1% (14.7-24.1)	-	-	-	-
Muehlen et al. (2003)	Balbino, Cascavel – State of Ceará	Cross-sectional study	548	51.3% (47.0-55.5)	-	-	-	-
Heukelbach et al. (2004b)	Balbino Cascavel – State of Ceará	Cross-sectional study	548	51.3% (47.0-55.5)	-	-	-	-
Heukelbach et al. (2004c)	Morro do Sandras, subarea Vicente Pinzón II, Fortaleza – State of Ceará	Cross-sectional study	Morro do Sandras: 849 Balbino: 505	Morro do Sandras: 54.4% (51.0-57.8) Balbino: 52.1% (47.6-56.5)	-	-	-	-

Heukelbach (2005)	Morro do Sandras, subarea Vicente Pinzón II, Fortaleza - State of Ceará	Cross-sectional study	1,185	March (2001): 33.6% (30.9-36.4) June (2001): 23.8% (21.4-26.3) September (2001): 54.4% (51.0-57.8) January (2002): 16.8% (14.4-19.4)	849	1,185	54.4% (51.0-57.8)	33.6% (30.9-36.4)
Muehlen et al., (2006)	Balbino, Cascavel – State of Ceará	Cross-sectional study	496	51.0%	-	-	-	-

Heukelbach et al. (2007)	Feliz Deserto – State of Alagoas	Cross-sectional study	Rainy season: 1,015 Dry season: 990	Rainy season: 21.6% (19.0-24.1) Dry season: 29.5% (26.6-32.3) p < 0.0001	990	1,015	29.5% (26.6-32.3)	21.6% (19.0-24.1)
Pilger et al. (2008a)	Balbino, Cascavel – State of Ceará Pedro de Souza, Cascavel – State of Ceará	Repeated cross-sectional study with intervention	Balbino; 459 Pedro de Souza: 140	Balbino: 37% (31-43) Pedro de Souza: 43% (35-51) P = 0.11	-	-	-	-
Pilger et al. (2008b)	Balbino, Cascavel – State of Ceará	Cross-sectional study	490	39% (34-43)	-	-	-	-
Damazio et al. (2009)	Criciúma – State of Santa Catarina	Cross-sectional study	917	1.6% (0.92-2.68)	-	-	-	-

Winter et al. (2009)	Morro do Sandras, Fortaleza – State of Ceará Balbino Cascavel – State of Ceará	Cross-sectional study	Morro do Sandras: 290 Balbino: 136	Morro do Sandras: 33.6% (30.9-36.4) Balbino: 51.1% (47.0-55.5)	-	-	-	-
Ariza et al. (2010)	Balbino, Cascavel – State of Ceará Feliz Deserto – State of Alagoas Morro do Sandras, Fortaleza – State of Ceará	Cross-sectional population-based study	7,121	21.1% to 54.4%	548 990 849	535 1,015 1,185	51.3% (47.1-55.5) 28.9% (26.1-31.7) 54.4% (51.1-57.8)	31.2% (27.3-35.2) 21.1% (18.6-23.6) 33.6% (30.9-36.3)
Bonfim et al. (2010)	Favela Wilma Maia, Natal – State of Rio Grande do Norte	Cross-sectional population-based study	310	23.2% (18.6-28.3)	-	-	-	-

% = percentage of infestation; CI = Confidence Interval; p = p value.

All studies had diagnostic confirmation through physical examinations performed by competent teams, except for one study, which obtained information through the application of questionnaires. Participants were referred to medical clinics or received treatment involving the extraction of the parasite and application or not of antibiotics in five studies; in one study participants were treated with either Ivermectin or Deltamethrin. The other studies (10) did not provide information regarding treatment.

The studied communities were all low-income. Levels of education were noted in almost all communities, with reported illiteracy rates ranging from 15% to 30%. The prevalence for this variable was even higher when incomplete primary education was considered together with illiteracy, reaching 75%.

Tungiasis prevalence considering seasonal variation

Variation between dry and rainy seasons was compared in three studies, which described higher prevalence of tungiasis in humans during dry seasons, due to factors associated with population and biological dynamics of parasite development, identified in Table 1.

Presence of animals as a risk factor for tungiasis

As the infestation by *T. penetrans* is anthropozoonotic, the presence of animals living in households and/or peridomiciles was noted in 11 of the 16 studies. Of these, dogs (11 studies) and cats (9) were most frequently reported, followed by pigs (5), horses (4), donkeys (3), cattle (2), chickens (2), rodents such as rats and mice (2), rabbits (1), goats (1) and monkeys (1). The prevalence of infestation was discussed in four studies, being higher in pet animals, such as dogs and cats, as can be seen in Table 2.

Assessment of the methodological quality of the included studies

Thirteen of the 16 titles were considered high quality studies, with total points above five; three studies obtained the lowest score with four points each. The average obtained in the quality assessment was 5.18, according to Table 3.

Table 2. Prevalence of infestation by *Tunga penetrans* in animals in households and peridomiciles, evaluated in scientific studies on the parasitosis in Brazil.

Authors and year	Animals	Prevalence
Carvalho et al. (2003)	Dogs	61%
Heukelbach et al. (2004c)		67.1%
Pilger et al. (2008b)		68.9%
Bomfim et al. (2010)		46.7%
Carvalho et al. (2003)	Cats	7.7%
Heukelbach et al. (2004c)		49.6%
Pilger et al. (2008b)		49.2%
Bomfim et al. (2010)		26.0%
Heukelbach et al. (2004c)	Rats	41.2%
Carvalho et al. (2003)	Pigs	16.6%
Heukelbach et al. (2004c)		0%
Pilger et al. (2008b)		0%
Carvalho et al. (2003)	Donkeys	0%
Heukelbach et al. (2004c)		0%
Pilger et al. (2008b)		0%
Heukelbach et al. (2004c)	Horses	0%
Pilger et al. (2008b)		0%
Heukelbach et al. (2004c)	Cattle	0%
Pilger et al. (2008b)		0%
Heukelbach et al. (2004c)	Monkeys	0%
Carvalho et al. (2003)	Chickens	0%
Heukelbach et al. (2004c)	Rabbits	0%
Heukelbach et al. (2004c)	Mice	0%

Table 3. Assessment of the methodological quality of scientific articles included in the systematic review: results obtained after using the Newcastle-Ottawa Scale for cross-sectional studies (NOS, 2009).

Authors and year	Criteria			
	Selection (0 to 5 points)	Comparability (0 to 2 points)	Outcome (0 to 3 points)	Total (0 to 10 points)
Heukelbach et al. (2002)	2	1	1	4
Wilcke et al. (2002)	3	1	1	5
Carvalho et al. (2003)	3	1	1	5
Heukelbach et al. (2003)	3	1	1	5
Muehlen et al. (2003)	4	1	1	6
Heukelbach et al. (2004b)	3	1	1	5
Heukelbach et al. (2004c)	3	1	1	5
Heukelbach (2005)	3	1	1	5
Muehlen et al. (2006)	4	2	1	7
Heukelbach et al. (2007)	4	1	1	6
Pilger et al. (2008a)	2	1	1	4
Pilger et al. (2008b)	4	1	1	6
Damazio et al. (2009)	3	1	1	5
Winter et al. (2009)	4	1	1	6
Ariza et al. (2010)	3	1	1	5
Bomfim et al. (2010)	2	1	1	4
Average				5.18

DISCUSSION

Tungiasis is an often-neglected health problem in poor communities in tropical and subtropical regions (Matias, 1989). Related to poverty, the disease presents several factors associated with infestations, such as poor sanitation, the presence of domestic and wild animals in the peridomicile and home and the prevalence of sandy soils, suitable for the development of the agent (Franck et al., 2003; Heukelbach, 2005; Heukelbach et al., 2005b). All these characteristics discussed in the literature were found and described in the communities analyzed in this systematic review, where the general prevalence found ranged from 1.6% to 54.8%, being higher in children in the 5-9 age group. To our knowledge, this is the first systematic review to address the issue in Brazil and its federative units.

The Favela de Serviluz and its surroundings, such as Morro do Sandras and the Vicente Pinzón subarea, municipality of Fortaleza, State of Ceará, was covered in seven of the analyzed studies (Table 1). Described as a poor region (Ariza et al., 2010), with approximately 60% of the population living on a monthly family income of less than two minimum wages (Heukelbach et al., 2002; Heukelbach et al., 2003; Heukelbach et al., 2004b; Heukelbach et al., 2005c). 30% of the adults presented high illiteracy rates (Wilcke et al., 2002), (Heukelbach et al., 2002; Heukelbach et al., 2003; Heukelbach et al., 2004b; Heukelbach et al., 2005c). The community is located close to the beach, where most homes had access to drinking water, but lacked sewage treatment and the municipal waste collection only served the neighboring areas, being insufficient for the population. In addition, part of the houses and streets had no paving and were in direct contact with the sand, favoring infestation (Heukelbach et al., 2002; Heukelbach et al., 2003; Heukelbach et al., 2004b; Heukelbach et al., 2005c).

The communities of Balbino and Pedro Souza are described respectively in seven and one of the studies (Table 1). Both are located in the municipality of Cascavel, State of Ceará, where floors were sandy and the nearest health center was located 10 km away (Pilger et al., 2008a). Located on the beach, in the Balbino community 8.4% of the houses were directly in contact with the sand, without flooring (Muehlen et al., 2003; Heukelbach et al., 2004b; Heukelbach et al., 2004c; Muehlen et al., 2006; Pilger et al., 2008a; Pilger et al., 2008b). Basic sanitation was considered inadequate, with garbage and sewage collection being insufficient for the entire population and 86.4% of houses having latrines (Heukelbach et al., 2004b). It is noteworthy that about 59.8% of the population lived below the poverty line, with a monthly per capita income of up to 60 Reais (Muehlen et al., 2003) with a 21% high school completion rate (Pilger et al., 2008b). These conditions are ideal for flea development, facilitating infestation in humans.

The community of Feliz Deserto, State of Alagoas, appears in two studies (Table 1), also characterized as low-income and with a high illiteracy rate among adults (Ariza et al., 2010). However, unlike the other communities studied, most households had cement floors and had garbage collected daily in the region (Heukelbach et al., 2007). In the literature, tungiasis has been reported as a parasitosis that mainly affects populations living in poverty (Muehlen et al., 2003), with high morbidity in poor urban and rural regions, with prevalence ranging from 16% to 54% (Bonfim et al., 2010).

The three remaining communities, Bananeiras (State of Rio de Janeiro) (Carvalho et al., 2003), Criciúma (State of Santa Catarina) (Damazio & Silva, 2009) and Favela Wilma Maia (State of Rio Grande do Norte) (Bonfim et al., 2010), were each described in one study (Table 1). All were reported as low-income and lacking basic sanitation and garbage collection services (Carvalho et al., 2003; Damazio & Silva, 2009; Bonfim et al., 2010). In Favela Wilma

Maia, 75% of the participants were illiterate or presented incomplete primary education (Bonfim et al., 2010) while 26.7% of household heads in Criciúma had not completed primary education (Damazio & Silva, 2009). Education is described in the literature as an indicator of socioeconomic status and health (Bonfim et al., 2010). Illiteracy and low education levels were variables identified in people with a parasite. These variables, together with housing conditions and precarious hygiene habits, are essential for the full development of citizenship and, when satisfactory, are essential to prevent the occurrence and spread of diseases (Bonfim et al., 2010).

The presence of animals was observed in all communities studied and is an important factor for the prevalence of infestation in humans, given the anthropozoonotic character of the disease. In Balbino (Pilger et al., 2008a) and Pedro Souza (Pilger et al., 2008b), the increase in the prevalence of infestation in humans was reportedly followed by the increase in infestation in animals, which continued to transmit and re-infect, even after treatment (Pilger et al., 2008a).

Free-living stages of *T. penetrans* develop generally in dry sandy soil (Heukelbach et al., 2005c; Heukelbach et al., 2007). Considering this characteristic of the agent's biological cycle, some of the analyzed studies (Heukelbach et al., 2005c; Heukelbach et al., 2007; Ariza et al., 2010) evaluated possible seasonal variations in the general prevalence of tungiasis in human populations, demonstrating a significant reduction in the rainy season in comparison with the dry period. Furthermore, it is possible to make a parallel between the characteristics of the soil favorable to the development of the agent and the fact that all the communities described, except for Criciúma (Damazio & Silva, 2009), presented unpaved houses and/or streets.

This systematic review conducts an unprecedented survey of the prevalence of tungiasis in Brazil. As a multifactorial parasitosis, infestation control is a challenge for health and zoonoses authorities, especially considering the neglected nature of the disease and its higher prevalence in children from low-income communities.

It is noteworthy, however, that this systematic review presents limitations, mainly in relation to finding and including studies that meet the proposed eligibility criteria. In spite of following standardized methodologies to avoid selection bias, there are numerous studies in the literature not published in indexed electronic databases. Considering that only the literature available in the electronic databases was reviewed and that the studies were chosen by the defined search strategies, the search in the gray literature was an alternative used to try to increase the number of sources and reduce possible selection bias. The studies included in this systematic review also showed heterogeneity. However, we tried to link similar aspects between them and zoonoses as much as possible in order not to exclude useful important information.

Although tungiasis is endemic in some Brazilian communities, there are few studies on its prevalence. In view of this, we encourage more researchers in the field of parasitology to conduct detailed studies on the epidemiological aspects of this neglected disease.

ACKNOWLEDGEMENTS

The authors would like to thank Mr. Gesner Xavier, a librarian from *Universidade Federal de Minas Gerais*, for his support during the definition of the descriptors and the search strategies.

CONFLICTS OF INTEREST.

The authors declare no conflicts of interest.

REFERENCES

1. Ariza L, Seidenschwang M, Buckendahl J, Gomide M, Feldmeier H, Heukelbach J. Tungiasis: a neglected disease-causing severe morbidity in a shantytown in Fortaleza, State of Ceara. *Rev Soc Bras Med Trop 40:* 63-67, 2007.
2. Ariza L, Wilcke T, Jackson A, Gomide M, Ugbomoikoto US, Feldmeier H, Heukelbach J. A simple method for rapid community assessment of tungiasis. *Trop Med Inter Health 15*: 856-864, 2010.
3. Bonfim MW, Cardoso DM, Cardoso VA, Andreazze R. Tungíase em uma área de aglomerado subnormal de Natal-RN: prevalência e fatores associados. *Epidemol Serv Saúde 19*: 379-388, 2010.
4. Carvalho RW, De Almeida AB, Barbosa-Silva SC, Amorim M, Ribeiro PC, Serra-Freire NM. The patterns of tungiasis in Araruama Township, State of Rio de Janeiro, Brazil. *Mem Inst Oswaldo Cruz 98:* 31-36, 2003.
5. Damazio OR, Silva MV. Tungiasis in schoolchildren in Criciuma, Santa Catarina State, South Brazil. *Rev Inst Med Trop São Paulo 51*: 103-108, 2009.
6. Eisele M, Van Marck E, Mehlhorn H, Meckes O, Franck S, Feldmeier H. Investigations on the biology, epidemiology, pathology and control of *Tunga penetrans* in Brazil: I. Natural history of tungiasis in man. *Parasitol Res 90:* 87-99, 2003.
7. Feldmeier H, Heukelbach J, Eisele M, Souza AQ, Barbosa LM, Carvalho CB. Bacterial superinfection in human tungiasis. *Trop Med Int Health 7*: 559-564, 2002.
8. Feldmeier H, Eisele M, Saboia-Moura R, Heukelbach J. Severe tungiasis in underprivileged communities: case series from Brazil. *Emerg Infect Dis 9*: 949-955, 2003.
9. Franck S, Feldmeier H, Heukelbach J. Tungiasis: more than an exotic nuisance. *Travel Med Infect 1:* 159-166, 2003.
10. Heukelbach J, Sales De Oliveira FA, Hesse G, Feldmeier H. Tungiasis: A neglected health problem of poor communities. *Trop Med Int Health 6*: 267-672, 2001.
11. Heukelbach J, Wilcke T, Eisele M, Feldmeier H. Ectopic localization of tungiasis. *Am J Trop Med Hyg 67:* 214-216, 2002.

12. Heukelbach J, Van Haeff E, Rump B, Wilcke T, Moura RC, Feldmeier H. Parasitic skin diseases: health-care seeking in a slum in northeast Brazil. *Trop Med Int Health 8*: 368-373, 2003.
13. Heukelbach J, Franck S, Feldmeier H. High attack rate of *Tunga penetrans* (Linnaeus 1785) infestation in an impoverished Brazilian community. *Trans R Soc Trop Med Hyg 98:* 431-434, 2004a.
14. Heukelbach J, Winter B, Wilcke T, Muehlen M, Albrecht S, Sales de Oliveira FA, Kerr-Pontes LRS, Liesenfeld O, Feldmeier H. Selective mass treatment with ivermectin to control intestinal helminthiases and parasitic skin diseases in a severely affected population. *Bull WHO 82*: 563-571, 2004b.
15. Heukelbach J, Costa AM, Wilcke T, Mencke N, Feldmeier H. The animal reservoir of *Tunga penetrans* in severely affected communities of northeast Brazil. *Med Vet Entomol 18*: 329-335, 2004c.
16. Heukelbach J. Tungiasis. *Rev Inst Med Trop de São Paulo 47*: 307-313, 2005.
17. Heukelbach, J, Walton, SF, Feldmeier H. Ectoparasitic infestations. *Current Infectious Disease Reports 7:* 373-380, 2005b.
18. Heukelbach J, Wilcke T, Harms G, Feldmeier H. Seasonal variation of tungiasis in an endemic community. *Am J Trop Med Hyg 72:* 145-149, 2005c.
19. Heukelbach J, Jackson A, Ariza L, Calheiros C, Soares V, Feldmeier H. Epidemiology and clinical aspects of tungiasis (sand flea infestation) in Alagoas state. *Brazil J Infect Dev Countries 1:* 202-209, 2007.
20. IBGE. Instituto Brasileiro de Geografia e Estatística. *Brasil: Cidades e Estados. 2021.* Available at: https://www.ibge.gov.br/cidades-e-estados.html?view=municipio. Accessed at: 13.jan.2021.
21. Linardi PM, Avelar DM, Filho EJF. Establishment of *Tunga trimamillata* (Siphonaptera: Tungidae) in Brazil. *Parasitol Res 112*: 3239-3242, 2013.
22. Linnaeus C von. *Systema naturae per regna tria naturae: secundum classes, ordines, genera, species, cum characteribus, differentiis, synonymis, locis.* 13ed. Aucta, Reformata: Amsterdam, 1758. 532p.
23. Luchini C, Stubbs B, Solmi M, Veronese N. Assessing the quality of studies in meta-analyses: Advantages and limitations of the Newcastle Ottawa Scale. *World J Meta-Anal 5*: 80-84, 2017.
24. Matias RS. Epidemics of tungiasis in Rio Grande do Sul. *Rev Soc Bras Med Trop 22*: 137-142, 1989.
25. Mazigo H, Bahemana E, Konje E, Dyegura O, Mnyone L, Kweka E, Kidenya BR, Heukelbach J. Jigger flea infestation (tungiasis) in rural western Tanzania: high prevalence and severe morbidity. *Trans R Soc Trop Med Hyg 106*: 259-263, 2012.
26. Methley AM, Campbell S, Chew-Graham C, McNally R, Cheraghi-Sohi S. PICO, PICOS and SPIDER: a comparison study of specificity and sensitivity in three search tools for qualitative systematic reviews. *Health Services Research 14*: 1-10, 2014.
27. Muehlen M, Heukelbach J, Wilcke T, Winter B, Mehlhorn H, Feldmeier H. Investigations on the biology, epidemiology, pathology and control of *Tunga penetrans* in Brazil: II. Prevalence, parasite load and topographic distribution of lesions in the population of a traditional fishing village. *Parasitol Res 90*: 449-455, 2003.
28. Muehlen M, Feldmeier H, Wilcke T, Winter B, Heukelbach J. Identifying risk factors for tungiasis and heavy infestation in a resource-poor community in Northeast Brazil. *Trans R Soc Trop Med Hyg 100:* 371-380, 2006.
29. Nair SKP, Zelalem TL, Tsehayneh K, Mehidi K, Ramaswamy V. Assessment of socioeconomic status and the prevalence of tungiasis in Jimma and Wolaita Sodo, Ethiopia. *Int J Intg Med Sci 3:* 211-215, 2016.

30. NEREUS. Núcleo de Economia Regional e Urbana do Estado de São Paulo. 2020. *Mapa do Estado do Maranhão*. Available at: http://www.usp.br/nereus/?dados=brasil. Accessed at: 13.jan.2021.

31. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, Shamseer L, Tetzlaff JM, Akl EA, Brennan SE, Chou R, Glanville J, Grimshaw JM, Hróbjartsson A, Lalu MM, Li T, Loder EW, Mayo-Wilson E, McDonald S, McGuinness LA, Stewart LA, Thomas J, Tricco AC, Welch VA, Whiting P, Moher D. The PRISMA 2020 statement: An updated guideline for reporting systematic reviews. *PLoS Med 18*: e1003583, 2021.

32. Pampiglione S, Fioravanti ML, Gustinelli A, Onore G, Mantovani B, Luchetti A, Trentini M. Sand flea (Tunda spp.) infections in humans and domestic animals: state of the art. *Med Vet Entomol 23*: 172-186, 2009.

33. Pilger D, Schwalfenberg S, Heukelbach J, Witt L, Mencke N, Khakban A, Feldmeier H. Controlling tungiasis in an impoverished community: An intervention stud. *Plos Negl Trop Dis 2:* e324, 2008a.

34. Pilger D, Schwalfenberg S, Heukelbach J, Witt L, Mehlhorn H, Mencke N, Khakban A, Feldmeier H. Investigations on the biology, epidemiology, pathology, and control of *Tunga penetrans* in Brazil: VII. The importance of animal reservoirs for human infestation. *Parasitol Res 102:* 875-808, 2008b.

35. PROSPERO. *International Prospective Register of Systematic Reviews. 2020.* Available at: https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=147957. Accessed at 13.jan.2021.

36. Ugbomoiko US, Ariza L, Ofoezie IE, Heukelbach J. Risk factors for Tungiasis in Nigeria: identification of targets for effective intervention. *PLoS Negl Trop Dis 1*: 87, 2007.

37. Wells GA, Shea B, O'Connell D, Peterson J, Welch V, Losos M, Tugwell T. *The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomized studies in meta-analyses.* 2009. Available at: http://www.ohri.ca/programs/clinical_epidemiology/oxford.asp. Accessed at 13.jan.2021.

38. Wilcke T, Heukelbach J, Moura RCS, Kerr-Pontes LRS, Feldmeier H. High prevalence of tungiasis in a poor neighbourhood in Fortaleza, Northeast Brazil. *Acta Trop 83*: 255-258, 2002.

39. Winter B, Oliveira FA, Wilcke T, Heukelbach J, Feldmeier H. Tungiasis-related knowledge and treatment practices in two endemic communities in northeast Brazil. *J Infect Dev Ctries 3:* 458-466, 2009.

40. Witt LH, Linardi PM, Meckes O, Schwalfenberg S, Ribeiro RA, Feldmeier H, Heukelbach J. Blood-feeding of *Tunga penetrans* males. *Med Vet Entomol 18*: 439-441, 2004.