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► **To cite this version:**

Sorya Belaz, Eugénie Gay, Florence Robert-Gangneux, Jean-Claude Beaucournu, Claude Guiguen. Tungiasis Outbreak in Travelers From Madagascar. *Journal of Travel Medicine*, 2015, 22 (4), pp.263-266. 10.1111/jtm.12217 . hal-01162380

HAL Id: hal-01162380

<https://hal-univ-rennes1.archives-ouvertes.fr/hal-01162380>

Submitted on 27 Oct 2016

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1 **BRIEF COMMUNICATION**

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3 **Tungiasis outbreak in travelers from Madagascar**

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13 **Running title:** Tungiasis outbreak in travelers from Madagascar

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26 **Abstract**

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28 Seven patients from a group of 16 travellers were diagnosed at our institution with one or

29 more sand fleas on toes, 1 day to 3 weeks after coming back from Madagascar. A

30 questionnaire was sent to the whole group to collect clinical and epidemiological information,

31 9/13 (69%) had received pre-travel medical advices, but none were aware of sand flea, thus

32 prevention measures were rarely applied. 5/7(71%) wore open sandals all over the trip.

33 Overall 10 sand fleas were extracted.

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53 Tungiasis is an endemic neglected disease in numerous tropical countries¹, due to the
54 penetration into the epidermis of a female sand flea laying its eggs. Although it is widespread
55 in local population, it is rarely reported in travellers. A survey of 165 travellers coming back
56 from tropics with dermatoses reported only 7 cases during 6 months of following up
57 travellers². Another study by Lederman *et al.* reported 31 diagnoses of tungiasis among 4742
58 dermatologic disorders diagnosed during a 10-year survey in worldwide travellers (in
59 GeoSentinel database)³. We report here a serie of 7 tungiasis cases from a group of 16 French
60 travellers coming back from Madagascar. Such grouped cases have been rarely described in
61 travellers, so we investigated contamination factors in the whole group, and we recall the
62 epidemiology of this ectoparasitose and prevention measures.

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64 **Case report**

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66 Sixteen travellers went backpacking for 14 days (3 nights in tents on the sand) along the
67 Pangalanes Channel (provinces of Antananarivo, Toamasina and Fianarantsoa, on the East
68 Coast of Madagascar), from November 10th to November 24th, 2012. Their average age was
69 61, with an equal number of men and women (Table 1). We've been referred the first patient
70 by his general practitioner one month after he returned home. He was complaining from two
71 "painful masses" under his fifth toe (Figure 1) which had appeared as soon as he came back.
72 Once he was diagnosed in our ward with tungiasis, he got in touch with his travelling
73 companions: four of them came at our consultation to have medical advice. One female
74 patient had noticed a warty lesion on her big toe that appeared as soon as she came back from
75 Madagascar, and decided to treat herself with cryotherapy (considered as self-exploration in

76 table 1). After being alerted by her past travelling companion, she came to our ward for
77 advice. Cryotherapy had indeed killed the flea, but the patient rather preferred the parasite to
78 be extracted. After further examination, another subungual flea was found on her 5th toe. The
79 three other patients who came to our ward reported painful toes 3 weeks after their return. All
80 were diagnosed with tungiasis. Two patients with painless lesions did not wish to receive any
81 medical treatment, and extracted the flea on their own. The pain depended on each patient
82 since it's just after being contacted by the first patient that they came for advice. Each patient
83 was examined in order to find other lesions. Fleas were extracted surgically, as described by
84 Pradinaud et al.¹. Overall, 10/12 fleas were extracted and identified by binocular examination
85 as *Tunga penetrans*. The outcome was unremarkable, except for two patients who suffered
86 from local infectious complication after extraction (one medical extraction in our ward and
87 one self-extraction with cryotherapy), which was resolved within 7 days under daily antiseptic
88 wash (Hibiscrub®). At the same time, we systematically checked the anti-*Tetanus* vaccination
89 status of each patient.

90 A questionnaire was sent to all 16 travellers to investigate the occurrence of similar clinical
91 signs in other person of the group and get information on prevention measures. Thirteen
92 people filled the questionnaire (response rate 81%). The attack rate was 53.8% (7/13). It
93 appeared that they walked rarely barefoot, except when swimming in Pangalanes Channel and
94 when crossing rivers. Infected patients were more likely to wear open sandals (5/7, 71%) than
95 healthy travellers (3/6, 50%) (Table 1). After clinical examination or from the questionnaire
96 answers, it appeared that three patients (43%) had a single lesion (3rd, 5th toe and foot sole
97 respectively); three patients (43%) had 2 lesions (1st and 5th toe for one patient, both 5th toes
98 for two patients) and one patient (14%) had 3 lesions (5th toes) (Table 1). The mean number of
99 fleas per patient was 1.7. Lesions were mainly observed on toes (11/12 fleas, 92%), and 41%

100 (5/12) of lesions were subungual. Clinical signs are described in Table 1. The mean time
101 between end of travel and onset of symptoms (itch or pain) was 15 days.

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103 **Discussion**

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105 Tungiasis is caused by the burrowing into the patient's epidermis of a female *Tunga*
106 *penetrans* (Siphonaptera) which only measures from 0.8 to 1 mm. In South America species
107 diagnosis is necessary because other species than *T. penetrans*⁴ can be found. Lesions are
108 mainly located on feet, especially on toes (with a common subungual localization), and rarely
109 reach the malleoli, probably because of the fleas' poor jumping ability. Most lesions observed
110 in our patients were located on toes (11/12 fleas, 92%) and mainly on the 5th toe (9/12 fleas,
111 75%). This localization on the 5th toe is not typical, as Veraldi et al.⁵ series of tungiasis,
112 found this localization in only 1/19 travellers(1/25 fleas) but can be explained by the fact this
113 toe often sticks out of flip-flops and sandals, as all patients with 5th toe lesion(s) did wear
114 open sandals (data not shown).

115 Due to feeding and egg production, the flea's abdomen extends. This phenomenon leads to a
116 whitish lesion, which measures from 5 to 7 mm, and is centered by a dark cutaneous roll
117 corresponding to the insect's excrements (Figure 1). Females shed about thousands eggs over
118 a 3- to 4-weeks period, after which they die. Eggs are visible to naked eyes⁶; and although
119 none of our patients saw them, we did see them when we extracted the fleas. Egg laying starts
120 within 8 to 10 days. Clinically, early pruritus (described by 5/7, 71% of our patients) is
121 quickly followed by an inflammatory pain (also described by 5/7, 71% of our patients) (Table
122 1). These typical lesions do not have any classic differential diagnoses, but can be often
123 unknown outside endemic areas, and can remain undiagnosed in travellers.

124 In our series, the free interval before the onset of symptoms seems longer (average 15 days,
125 median 21 days) than usually described in other series. Caumes *et al.*⁷ described a median
126 interval of only 12 days. In another series of 9 cases in travellers back from Ethiopia, Grupper
127 *et al.*⁸ described an interval between contamination and the observation of visible lesion of 7
128 to 12 days. This longer interval observed in our series can be explained by the fact that most
129 patients did not notice the lesion before the onset of symptoms.

130 *T. penetrans* was exclusively present in Central and South America, as well as in the
131 Caribbean, until it was imported to Sub-Saharan Africa, apparently in the eighteenth century⁶.
132 Tungiasis has not yet been established in Europe and USA, despite so many occasional
133 records⁹. Since then, it spread across the region (especially during the nineteenth century) and
134 finally reached Madagascar. From 1950 to 1960, indoor spraying campaigns targeting malaria
135 vectors in Madagascar probably had an impact on reducing the importance of this parasite in
136 the island. However, local populations still remain highly exposed to sand fleas, especially in
137 districts with hog farms, since pigs are hosts for *T. penetrans* as well¹⁰. Two of the coastal
138 zones along the Pangalanes Channel (visited by our travellers) are among the most infested
139 areas with *T. penetrans*. The active period of the parasite peaks during the dry season¹¹, which
140 lasts in Madagascar from May to November. Our patients were there in November at the end
141 of the high risk season, which can explain the high attack rate.

142 Travellers usually have fewer sand fleas compared to local population, probably due to a
143 shorter duration of exposure. Complications usually remain local, and mainly occur when
144 patients manipulate their own lesions, as described by Veraldi *et al.*⁵. In our series, 2/4 (50%)
145 patients who manipulated their lesions developed superinfection.

146 The pre-travel consultation allows to inform travellers about sand fleas (especially in case of
147 travel in endemic areas during dry season). Nine patients (69%) benefited from a pre-travel
148 medical advice but none of them have had advice about sand fleas (Table 1). The knowledge

149 of the disease enables a quick treatment, and helps avoiding complications. Personal
150 prevention among travellers is achieved through wearing closed shoes but seems rather utopic
151 in tropical zones. It also relies on frequent applications of diethyltoluamine-based skin
152 repellents on feet, a prophylaxis which may be easier to follow, even in wet and warm areas.
153 In the present report, most contaminated patients wore open shoes (5/7, 71%) and none used
154 repellent on their feet (0/7, 0%) (Table 1). Some healthy travellers wore closed shoes (3/6,
155 50%) and one used repellent on his feet (1/6, 17%), but the comparison between infected and
156 non-infected patients did not reveal statistically significant results, probably due to a lack of
157 power.

158 Whether tungiasis is not an important burden in the whole population of travellers^{2,3}, it must
159 be taken into consideration for backpackers who are particularly at risk in endemic areas. This
160 report describes a large outbreak of tungiasis in travellers. Although Grupper *et al.* already
161 described a similar outbreak (9 cases), we medically examined more patients (5/7 patients)
162 and the interest of our study is that most travellers of the group (13/16), whether infected or
163 not, responded to a questionnaire, so that hypotheses on contamination can be discussed.
164 This report shows the importance of pre-travel medical consultation as an opportunity to
165 inform travellers and provide prophylaxis counseling for stays in countries at risk.

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168 **Declaration of Interests**

169 The authors state they have no conflicts of interest to declare.

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202 **Table 1:** Demographic and clinical characteristics of the 13/16 responding travellers

	Travellers with sand flea(s) (N=7)	Travellers without sand flea (N=6)	All (N=13)	203
Mean age, y [min-max]	60.6 [55-67]	62.6 [60-65]	61.3 [55-67]	204
Sex, M/F	4/3	3/3	7/6	205
Pre travel advises, n/N (%)	4/7 (57%)	5/6 (83%)	9/13 (69%)	207
Advises on sand fleas	None	None	None	208
Use of skin repellent on feet, n/N (%)	None ^a	1/6 (17%)	1/13 (8%)	209
Walking barefoot, n/N (%)	7/7 (100%)	6/6 (100%)	13/13 (100%)	210
Wearing open sandals, n/N (%)	5/7 (71%)	3/6 (50%)	8/13 (62%)	211
Visiting hog farm	None	None	None	212
Contact with stray dogs	None	None	None	213
Skin lesions, n/N (%)				214
1 lesion	3/7 (43%)	na	na	215
2 lesions	3/7 (43%)	na	na	216
3 lesions	1/7 (14%)	na	na	217
Clinical signs, n/N (%)				218
Itch	5/7 (71%)	na	na	219
Pain	5/7 (71%)	na	na	220
Super infection	2/7 (29%)	na	na	221
Mean time return-lesions, days [min-max]	15 [0-21]	na	na	222
Self-exploration, n/N (%)	4/7 (57%)	na	na	
Medical exploration, n/N (%)	5/7 (71%)	na	na	

223 ns: non-significant, na: not applicable

224 ^a 4 travellers with sand fleas used repellent but not on feet

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229 **Figure 1:** Single lesion of the right fifth toe. Typical 'mistletoe' centered lesion with a black

230 ring

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