

Case Report and Literature Review: Podoconiosis in Southwestern Uganda

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AUTHORS' CONTRIBUTIONS

This work was carried out in collaboration between all authors. [Author Philip Dwek](#) wrote the preliminary manuscript with help from [Meghan Wafer](#), who [also reviewed background information and wrote](#) ~~wrote and extensively reviewed~~ the ~~topic background~~ introduction. [Authors William Cherniak](#) and [Michael Silverman](#) made multiple edits and suggestions. [Ling Yuan Kong](#) visited local villages to find the second case. All authors read and approved the final manuscript.

ABSTRACT

Background/Aims: This report presents one confirmed and two suspected cases of podoconiosis in the Kabale region of southwestern Uganda. Podoconiosis has not previously been described ~~in Uganda in Uganda~~ outside of the ~~e~~Eastern region bordering Kenya. [The aim of this case report is to](#) ~~increase awareness of the condition~~ [can help in in order to](#) ~~enhance~~ prevention and treatment efforts.

Presentation of Cases: Podoconiosis ~~is a debilitating disease resulting in asymmetrical ascending bilateral lymphedema of the feet and legs.~~ It is associated with chronic barefoot exposure to red volcanic soil, [with greater prevalence in high-altitude in](#), impoverished areas of the tropics.

~~We~~ [This report](#) describes ~~one case and two suspected cases of podoconiosis in south western Uganda one confirmed case and two suspected cases.~~ Case 1 was confirmed by a negative filarial antigen detection test. Cases 2 and 3 had negative filarial smears, but antigen detection kits were not available onsite at the time. However, the altitude at which all three patients live (>1500 masl) makes filarial disease unlikely.

Discussion: Podoconiosis has not been previously reported in the region, and thus may be under-diagnosed due to a low index of suspicion among clinicians. Using adequate footwear

is an important component of prevention. However, limited financial resources present a significant barrier to the use of footwear. Understanding community attitudes towards causes and risk factors is also integral to effective prevention.

Conclusion: Podoconiosis has a high potential for elimination, as it is preventable and treatable in the early stages with consistent use of footwear and regular foot-washing. In addition, increased awareness among physicians and clinicians of the presence of the disease in this area can lead to early detection and treatment.

Keywords: Podoconiosis, Uganda, mossy foot, case report

1. INTRODUCTION

Podoconiosis, or non-filarial elephantiasis, is a non-infectious disease characterized by ascending bilateral and asymmetrical lymphedema of the feet and legs, generally occurring below the knees. It is associated with chronic barefoot exposure to volcanic soil. Local inflammation is thought to be caused by an abnormal inflammatory reaction to irritant mineral particles in red volcanic clay ⁽¹⁾ [1], which may lead to fibrosis and obstruction of lymphatic vessels. The condition is occasionally accompanied by the appearance of “mossy” nodules on the skin. Podoconiosis is a debilitating disease, with a usual onset in the third decade of life ⁽²⁾ [2]. A major and serious complication of podoconiosis is acute adenolymphangitis (ALA), which presents as a warm, painful sensation in the limbs, accompanied by fever. Diagnosis of podoconiosis is usually made based on patient history and clinical examination, and is supported by a negative filarial antigen test ⁽³⁾ [3]. ⁽³⁾

~~Podoconiosis is found in tropical areas, notably Africa.~~ Podoconiosis has also been documented in ~~tropical areas worldwide, most notably in Central America and Asia, particularly in India and Africa~~ [4]. ⁽⁴⁾ In Ethiopia, the average prevalence of podoconiosis in endemic red clay soil areas is approximately 5%, making it a significant public health problem ⁽⁵⁻⁶⁾ [5-6]. ~~(5-7)(5-6)~~ Increased prevalence has been noted at altitudes of 1500 meters above sea level (masl) or higher, in areas that receive an annual rainfall $\geq$ 1500 mm ~~and with those that have~~ an average annual temperature between 19-21°C ⁽⁴⁾ [4]. The condition is prevalent among farmers and workers of lower socioeconomic status, especially those who work barefoot. Avoiding barefoot work, along with regular foot-washing, has been shown to decrease progression of the disease ⁽⁷⁾ [7]. ~~(8)(7)~~ Unfortunately, the individuals typically affected often work in agricultural settings earning a subsistence income, and thus cannot afford appropriate footwear.

In addition to the environmental risk factors outlined above, genetic factors, including HLA associations, have been linked to increased risk ⁽⁸⁾ [8]. ~~(9)(8)~~ ~~Because of the genetic factors~~ Thus, it is not uncommon for affected individuals to have a family history of podoconiosis and for there to be increased risk among siblings. This familial clustering ~~of podoconiosis~~ due to a common genetic background is accentuated by the clustering of poverty (another risk factor) within families. The potential heritability of the condition and resultant physical disability leads to further social isolation, particularly with reduced marriage opportunities. In one study conducted in ~~s~~ Southern Ethiopia ⁽⁹⁾ [9]. ~~(10)(11)~~ 30% of individuals affected felt they were forced to dissolve marriage plans because of the condition.

In Uganda, podoconiosis has not been previously described outside of the eastern region around Mt. Elgon, bordering Kenya ⁽¹⁰⁾ [10]. ~~(11)(12)~~ However, in the remote past Lowenthal ~~in 1934~~ ⁽¹¹⁾ [11] ~~(12)(13)~~ identified it in the capital city, Kampala, ~~in the remote past~~, but at that time

confirmation of the disease by antigen detection testing was not possible. ~~Podoconiosis has been found~~ in Rwanda, which borders Uganda to the southwest. ~~In Rwanda the prevalence of podoconiosis~~ has previously been estimated at 0.6% [1]. (1) In this paper, we describe one confirmed case and two other suspected cases of podoconiosis in the Kabale region of southwestern Uganda, near the Rwandan border. This is an impoverished rural area, populated primarily by subsistence farmers, which has with fertile red volcanic soil ~~and is populated primarily by subsistence farmers.~~

2. PRESENTATION OF CASES

Patients were seen during mobile rural medical outreach clinics in the Kabale region. All patients gave verbal consent for publication of the cases and photos. ~~The first case~~ Case 1 was seen in March 2013; Cases 2 and 3 were seen in February 2014.

2.1 Case 1

A 72-year-old Ugandan male presented to a mobile medical and dental clinic in Muyumbu village with a chronic history of bilateral swelling of the feet and legs (Fig. 1). The swelling had developed slowly over 15 years, starting at age 57. He had been working barefoot for most of his life as a subsistence farmer, and reported episodes (which would resolve spontaneously) of increased swelling associated with tenderness ~~that would resolve spontaneously~~. There was no history of hypertension or diabetes and no clinical evidence of congestive heart failure. He was a widower with seven children, with no affected family members.



Fig. 1. Patient 1: Bilateral lower limb swelling ~~observed in patient 1.~~

Upon examination, bilateral swelling of the legs, hyperkeratosis of the dorsal and plantar aspects of both feet (Fig. 2), and moss-like papillomata were discovered. Sensation was normal to pinprick and light touch. The patient reported feeling socially isolated because of his legs. He was unable to work, as he required crutches to ambulate. A filarial antigen detection test (BinaxNOW Filariasis, Alere. Providence, Rhode Island) was negative.



Fig. 2. Patient 1: Hyperkeratosis on dorsal right foot-of patient 1.

2.2 Case 2

A 70-year-old married woman with an approximate 40-year history of bilateral lower limb edema ([Fig. 3](#)) was discovered in her local village. The patient was from Bubare subcounty (1800 masl) but had been living in Nyakabungo (2019 masl) for many years. She first noticed her symptoms during pregnancy at the age of 30. She worked as a farmer, with no formal education, and reported never having worn footwear. She had experienced occasional episodes of suspected ALA, up to three times per year.

~~She~~ The patient reported feeling stigmatized, and had worried for many years that her children would not be able to marry should they develop symptoms of the disease. ~~Fortunately all of her children remained disease-free and were subsequently married. The patient~~ She reported that her mother had shown similar symptoms ~~and thus may also have had pedoconiosis~~. There were no discrete lesions noted; sensation was not assessed, as leprosy was not suspected.



Fig. 3. Case Patient 2 with Bilateral Lymphedema and webbing of the toes.

2.3 Case 3

A 70-year-old male farmer presented to a mobile medical outreach clinic with a history of bilateral leg edema starting in his teenage years. He was from Kashekye village (1632 masl) and had never left Uganda, nor had he traveled within the country, except for a six-month period spent in Bunyoro (in western Uganda). He reported that his father had been afflicted with similar symptoms. Of his 14 children, he believed that one son had developed the condition. However, this son was not available for examination.

The patient denied ever feeling stigmatized or isolated from other members of the community. He consistently went barefoot, unless he was attending church or another community gathering. Although he denied experiencing symptoms/signs of ALA, he admitted to spending approximately two days per month in bed due to his condition. Sensation to pinprick and light touch were normal.

3. DISCUSSION

We present one confirmed and two suspected cases of podoconiosis in southwestern Uganda. Case 1 was confirmed by a negative filarial antigen detection test. Blood smears taken in Cases 2 and 3 to rule out filariasis as a possibility were negative. (although these were taken during the day) rather than at midnight, which would have improved sensitivity to filariasis detection. and had no eosinophilia observed on blood smears. Both individuals lived at altitudes greater than 1500 masl, making filariasis unlikely. However, Filarial antigen detection kits were not available onsite in 2014. Therefore, Cases 2 and 3 are referred to as suspected cases rather than confirmed ones.

Despite the unavailability of cases 2 and 3 in order to rule out filariasis as a possibility. Antigen testing was not performed, as it was not available onsite at the time. Furthermore, blood samples were drawn during the day rather than at midnight, which would have improved sensitivity to filariasis detection. Despite this, we believe that the altitude at which all three cases live (>1500 masl) makes filarial disease unlikely. An assessment of lymphatic

166 filariasis in Uganda, made by screening 17,533 children, found no circulating filarial antigens
167 by immunochromatographic card tests above 1300 masl [12]. ~~(13)(15)~~ Eosinophilia was not
168 observed in either of the two suspected cases ~~discussed in this paper, which~~ further
169 suggestings that ~~their~~ these patients' conditions were due to podoconiosis rather than
170 filariasis. ~~Two of the three Ppatients 1 and 3s~~ had intact sensation to light touch and pinprick,
171 suggesting that leprosy was an unlikely ~~to be the~~ cause of their symptoms. Case Patient #2
172 ~~(who was listed as aone of the~~ suspected cases) was not assessed for neuropathy.

173
174 Several patients attending the clinic reported having "seen this before," suggesting the
175 possible presence of additional cases of podoconiosis in this region. As house-to-house
176 surveys for the disease were not carried out, we cannot estimate the local prevalence rate.
177 The number of patients reporting to clinics is likely an under-representation of the true
178 prevalence because of the difficulty in attending clinics for patients with mobility issues
179 caused by the disease. ~~As well, S~~some patients may also actively avoid outsiders because
180 of the stigma associated with a diagnosis of podoconiosis and thus not present to clinic [13-
181 14].(10,14)

182
183 It has been found that a simple management regimen of education on the etiology of the
184 disease, along with prevention—including foot hygiene, use of emollients, elevation of limbs
185 at night, and the wearing of shoes and socks—is effective in improving both clinical
186 outcomes ~~as well asand~~ the dermatological quality of life index (DLQI). This simple approach
187 can lead to a decrease in mean leg circumference by 2 cm after one year and a change in
188 mean DLQI to a level where the condition minimally affects daily life [7].(8)(7) Unfortunately,
189 individuals typically affected by podoconiosis often work in agricultural settings earning a
190 subsistence income and cannot afford appropriate footwear.

191
192 A 2013 study conducted by Ayode et al [15(14)(16)] aimed to determine the barriers to
193 wearing footwear for Ethiopians living in rural areas. One major barrier was limited financial
194 resources. Large families could not afford shoes for each child, or would buy cheaper shoes
195 that did not provide adequate protection. Family members would also wear shoes only
196 occasionally in order to make them last longer. As well, some shoes were not suitable for all
197 activities: plastic or foam shoes were slippery or would allow mud to enter, and would thus
198 become heavy during farming. Individuals with established podoconiosis also found it difficult
199 to find shoes to fit swollen feet. Finally, some families without a history of podoconiosis
200 tended not to wear shoes because they believed the disease to be solely heritable and thus
201 ~~did not~~ believed themselves not to be at risk—.

202
203 On the other hand, the same study [15] also found that footwear is favored by the perception
204 that it is shameful to appear barefoot in public places, as well as by children's desire to wear
205 shoes to school. Programs such as the Mossy Foot Treatment and Prevention Program
206 Association (MFTPA) have favorably changed attitudes towards footwear [16].(15)(17)
207 Knowledge of which families have been affected- can potentially help identify which children
208 are at greater risk, in order to more efficiently allocate so that scarce resources for footwear
209 ~~can be allocated more efficiently~~ [17].(16)(18)

210
211 Understanding community attitudes towards the causes of podoconiosis and understanding
212 factors that increase risk are integral to prevention. Avode et al, who surveyed both affected
213 and unaffected adults in southern Ethiopia, found three common attitudes regarding the
214 cause of podoconiosis: (1) that the condition is solely heritable, (2) that genetics do not play
215 a factor, and (3) that the condition actually is caused by a mix of genetic and environmental
216 factors [15, 18].(14,17) (as is actually the case) [15, 18].(16,19) Those who believed
217 podoconiosis to be strictly heritable did not endorse shoe use, especially if there was no
218 family history. Those who did not think that genetics were involved believed shoes to be

important, but many also erroneously believed the condition to be contagious and thus avoided contact with the affected as a means of prevention. It should be noted that more unaffected than affected individuals were included in this study, which possibly skews the results. In addition, many

~~In Ethiopia, although a large majority of~~ health care professionals in Ethiopia, are while aware of ~~the condition~~the condition, ~~many~~ hold misconceptions about ~~the disease~~podoconiosis, specifically ~~the misconception that the disease~~it has an infectious component [19]. ~~(19)(20)~~

Podoconiosis has not previously been reported in southwestern Uganda, and thus the condition may be under-diagnosed due to a low index of suspicion among clinicians. ~~Local~~ Ppatients and physicians in southwestern Uganda were not aware of any ~~local~~ term for podoconiosis in the local ~~language of Kiga~~ language (also known as Ruchiga), likely reflecting the lack of ~~local~~ awareness of the disease. On the other hand, leprosy is recognized locally and is referred to as “ebibembe”, ~~and while~~ and while filariasis is referred to as “emirumo”.

4. CONCLUSION

Podoconiosis- is a preventable disease that is treatable in the early stages if shoes are worn consistently and if feet are washed regularly [7(8)](7). Therefore, podoconiosis has a high potential for elimination. Many individuals in rural Uganda are unable to afford shoes, which can lead to the progression of disability and a decrease in productivity and earning potential. The economic burden of podoconiosis is considerable. It is estimated that affected individuals lose 45% of their working days per year [20(19)-(21)].

This paper presents the first published case of podoconiosis in southwestern Uganda, as well as two other suspected cases. ~~The-. These cases are presented with the aim of information provided by these cases has the potential to raising~~ These cases are presented with the aim of increasing awareness of the condition in the region and ~~lead to~~increasing prevention efforts in populations at risk through adequate shoe use and foot-washing. In addition, increased awareness among physicians and clinicians of the presence of the disease in this area can lead to early detection and treatment, which is essential to decreasing disability and loss of productivity.

CONSENT

All authors declare that verbal informed consent was obtained from the patients for publication of this case report and accompanying images.

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COMPETING INTERESTS

The authors have no competing interests.

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