

Case Report and Literature review: Podoconiosis in South Western Uganda

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AUTHORS' CONTRIBUTIONS

This work was carried out in collaboration between all authors. Author PD wrote the preliminary manuscript with help from MW who also wrote and extensively reviewed the topic background. Authors WC and MS made multiple edits and suggestions. LYK visited local villages to find the second case. All authors read and approved the final manuscript.

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ABSTRACT

Background

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 in impoverished areas of the tropics.

Presentation of Case

We describe one case and two suspected cases of podoconiosis in south western Uganda.

Conclusion

Podoconiosis has not previously been described in Uganda outside of the Eastern region bordering Kenya. Increased awareness of the condition can help in prevention and treatment efforts and as well can help reduce stigma.

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), which may lead to fibrosis and obstruction of lymphatic vessels. The condition is occasionally accompanied by the appearance of "mossy" nodules on the skin; it is a debilitating disease, with a usual

onset in the third decade of life(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).

~~Although podoconiosis is found in tropical areas, notably Africa, it has also been documented in Central America and Asia particularly in India(4). In Ethiopia, the average prevalence of podoconiosis in endemic red clay soil areas is approximately 5%, the prevalence of podoconiosis has been estimated at ~5% making it a significant public health burden(5-7). In contrast, Rwanda has a reported prevalence of 0.6%(1).~~ Increased prevalence has been noted at altitudes above 1500 meters above sea level (masl), in areas that receive an annual rainfall $\geq$ 1500 mm and a temperature between 19-21°C(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(8). 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(9). Due to the genetic factors, as well as clustering of the risk factor of poverty within families, it is not uncommon for affected individuals to have a family history of podoconiosis and for there to be increased risk among siblings.

~~Not only is the disease itself debilitating,~~ 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 Southern Ethiopia, 30% of individuals affected felt they were forced to dissolve marriage plans because of the condition(12).

In Uganda, podoconiosis has not been previously described outside of the eastern region, around Mt. Elgon bordering Kenya (13). While there have been no recent reports of the disease outside the eastern part of the country, the original observations made by Lowenthal in 1934 suggest its occurrence in the capital city, Kampala(14). Prevalence in Rwanda (southwest border of Uganda) has previously been estimated at 0.6%(1). In this paper, we describe one confirmed case and two other suspected cases of podoconiosis in the Kabale region of southwestern Uganda, an impoverished rural area, with fertile red volcanic soil.

2. CASE PRESENTATION

Patients were seen during mobile rural medical outreach clinics in the Kabale region. The first case was seen in March 2013; Cases 2 and 3 were seen in February 2014.

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. The swelling 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 of increasing 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 7 children, with no affected family members. Upon examination, bilateral swelling of the legs, hyperkeratosis of the dorsal and plantar aspects of both feet, and moss-like papillomata were discovered. Sensation was normal to pinprick and light touch. The patient reported feeling socially isolated because of his legs and because he was unable to work, as he required crutches to ambulate. A filarial antigen test (BinaxNOW Filariasis, Alere. Providence, Rhode Island) was negative.



Figure 1. Bilateral lower limb swelling observed in patient 1.



Figure 2. Hyperkeratosis on dorsal right foot of patient 1.

Case 2

A 70-year-old married woman with an approximate 40-year history of bilateral lower limb edema 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 reported feeling stigmatized, and worried for many years that her children would not be able to marry should they develop podoconiosis. It should be noted, however, that all of her children remained disease-free and subsequently married. The patient had a positive family history of suspected podoconiosis in her mother. There were no discrete lesions noted, sensation was not assessed as leprosy was not suspected.



Figure 3: Case 2 with Bilateral Lymphedema and webbing of the toes.

Case 3

A 70-year-old male farmer presented to a mobile medical outreach clinic with bilateral leg edema, which he had been experiencing since 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 6-month period spent in Bunyoro (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, but the 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 at 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 was normal.

Note: Both Cases 2 and 3 had negative filarial smears (although these were taken during the day) 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 in 2014, Therefore, Cases 2 and 3 are referred to as suspected cases rather than confirmed ones.

3. DISCUSSION

In this paper, we report one case and two suspected cases of podoconiosis in southwestern Uganda. Several patients attending the clinic reported having “seen this before,” suggesting the presence of additional cases in this region. As house-to-house surveys for the disease were not carried out, we cannot estimate the local prevalence rate. The number of patients reporting to clinics is likely an under-representation of the true prevalence because of the difficulty in attending clinics for patients with mobility issues caused by the disease; as well, some patients are likely to actively avoid outsiders because of the stigma associated with a diagnosis of podoconiosis(11,15).

Blood smears were taken in the two suspected cases 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. Despite this, we believe that the altitude at which all three cases live (>1500 masl) makes filarial disease unlikely. An assessment of lymphatic filariasis in Uganda, made by screening 17,533 children, found no circulating filarial antigens by immunochromatographic card tests above 1300 masl(16). Eosinophilia was not observed in either of the two suspected cases discussed in this paper, which further suggests that their conditions were due to podoconiosis rather than filariasis. Two of the three patients had intact sensation to light touch and pinprick, making an alternative consideration; leprosy unlikely. The third patient (who was listed as a suspected case) was not assessed for neuropathy.

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

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 143 A 2013 study conducted by Ayode et al aimed to determine the barriers to wearing protective footwear faced by
 144 Ethiopians in rural areas(17). One major barrier was limited financial resources; large families could not afford shoes for
 145 each child and would wear shoes only occasionally in order to make them last longer, or would buy cheaper shoes that
 146 did not provide adequate protection. As well, some shoes were not suitable for all activities, as plastic or foam shoes were
 147 slippery or would allow mud to enter and become heavy during farming. Individuals with established podoconiosis also
 148 found it difficult to find shoes to fit swollen feet. Finally, some families without a history of podoconiosis tended not to wear
 149 shoes because they believed the disease to be solely heritable and thus did not believe themselves to be at risk. On the
 150 other hand, the same study also found that footwear is favored by the perception that it is shameful to appear barefoot in
 151 public places and by children's desire to wear shoes to school. Programs such as the Mossy Foot Treatment and
 152 Prevention Program (MFTPA) have favorably changed attitudes towards footwear(18). Potentially, family health history
 153 can help identify children at greater risk in order to allocate resources more efficiently(19).

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 155 Understanding community attitudes towards the causes of podoconiosis and understanding factors that increase risk are
 156 integral to prevention. Avode et al, who surveyed both affected and unaffected adults in southern Ethiopia found three
 157 common attitudes regarding the cause of podoconiosis: (1) that the condition is solely heritable, (2) that genetics do not
 158 play a factor, and (3) some local residents were aware that the condition actually is caused by a mix of genetic and
 159 environmental factors (17,20). Those who believed podoconiosis to be strictly heritable did not endorse shoe use,
 160 especially if there was no family history. Those who did not think that genetics were involved believed shoes to be
 161 important, but many also erroneously believed the condition to be contagious and thus avoided contact with the affected
 162 as a means of prevention. It should be noted that more unaffected than affected individuals were included in this study,
 163 which possibly skews the results.

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 165 In Ethiopia, although a large majority of health care professionals are aware of the condition, many hold misconceptions
 166 about the disease, specifically the misconception that the disease has an infectious component (21). Podoconiosis has
 167 not previously been reported in southwestern Uganda, and thus the condition may be under-diagnosed due to a low index
 168 of suspicion among clinicians. Local patients and physicians in South Western Uganda were not aware of any local term
 169 for podoconiosis likely reflecting the lack of awareness of the disease, but in the local language of Kiga (also known as
 170 Rukiga) leprosy is referred to as “ebibembe”, and filariasis is referred to as “emirumo”.

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 172
 173 Podoconiosis is a neglected tropical disease that has a high potential for elimination, as it is preventable and treatable in
 174 the early stages if shoes are consistently worn and feet are washed regularly(8). ~~Unfortunately, Mm~~ many individuals in rural
 175 Uganda are often unable to afford shoes, which could lead to progression of disability and a decrease in productivity and
 176 earning potential. The economic burden of podoconiosis is considerable, and it is estimated that affected individuals lose
 177 45% of their working days per year (22).

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4. CONCLUSION

This paper presents the first published case of podoconiosis in southwestern Uganda, as well as two suspected cases. The information provided by these cases has the potential to raise awareness of the condition and lead to prevention 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.

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COMPETING INTERESTS

The authors have no competing interests.

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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