

Case Report and Literature review: Podoconiosis in South Western Uganda

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 been documented mainly in two other continents; Central America and Asia particularly India[4]. In Ethiopia, the prevalence of podoconiosis has been estimated at ~5% making it a considerable disease 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 ≥ 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]. Therefore, 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, but individuals affected by podoconiosis also experience social stigma and isolation [10], related to the common misconception that the condition is contagious (while the reality is that it is generally associated with poverty) [11]. 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 bordering Kenya, in the area around Mt. Elgon [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]. In this paper, we describe one confirmed case and two other suspected cases of podoconiosis in Kabale, an impoverished rural region, with fertile red volcanic soil, in southwestern Uganda.

2. CASE PRESENTATION

Patients were seen during mobile rural medical outreach clinics in the Kabale region of southwestern Uganda. 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 in the feet and legs. 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. The swelling developed slowly over 15 years. 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. 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 Bubaresub county (1800 masl) but had been living in Nyakabungo (2019 masl) for many years. She noticed her symptoms during pregnancy around 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.



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 through 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, the patient 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.

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 [10,15].

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.

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

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

Understanding community attitudes towards the causes of podoconiosis and understanding factors that increase risk are integral to prevention. Avode et al, who surveyed both affected and unaffected adults in southern Ethiopia found three common attitudes regarding the cause of podoconiosis: (1) that the condition is solely heritable, (2) that genetics do not play a factor, and (3) that the condition is caused by a mix of genetic and environmental factors [17]. Those who believed podoconiosis to be strictly heritable did not endorse shoe use, especially if there was no family history. Those who did not think that genetics were involved believed shoes to be important, but many also 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 Ethiopia, although a large majority of health care professionals are aware of the condition, many hold misconceptions about the disease, specifically the misconception that the disease has an infectious component [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.

Podoconiosis is a neglected tropical disease that has a high potential for elimination, as it is easily preventable and treatable in the early stages if shoes are consistently worn and feet are washed regularly [8]. Unfortunately, many individuals in rural Uganda are often unable to afford shoes, which could lead to progression of disability and a decrease in productivity and earning potential. The economic burden of podoconiosis is considerable, and it is estimated that affected individuals lose 45% of their working days per year [21].

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.

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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