

Podoconiosis or non-filarial lymphedema: A case report from Kinshasa, Republic Democratic of Congo

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Abstract

Podoconiosis is considered one of the most stigmatizing health problem in endemic areas. The absence of adequate care leads to serious social and economic consequences. Here, we report the case of a patient who presented to the National Institute of Biomedical Research in Kinshasa (the Democratic Republic of Congo, DRC) for diagnosis confirmation of *Mycobacterium ulcerans* infection. A 55-year-old female patient, living for more than 10 years in the mining territory of Kahemba in the province of Kwango in the DRC, presented on inspection with thickened skin and papillomatous growths on the sides of the feet and heels. Swelling of the lower limbs was bilateral and asymmetric, and limited to the area just below the knees. There were also nodules and maceration between the toes. Tests for *Mycobacterium ulcerans* and microfilariae were negative. She was then referred to University Clinics of Kinshasa for further medical care. Although podoconiosis is a completely preventable disease, it remains poorly understood by many clinical practitioners and policy makers. Due to the lack of a standardized protocol and diagnostic tools, late discovery of non-filarial lymphedema sometimes leads to disabling consequences, as was the case for the patient in the present study.

Keywords: Podoconiosis, case report, non-filarial lymphedema, elephantiasis, Democratic Republic of Congo.

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I. Introduction

Podoconiosis, also known as endemic non-filarial elephantiasis, is a non-contagious, immune-mediated tropical disease caused by heavy metals affecting the lymphatic vessels. It is a form of non-filarial lymphedema resulting in chronic, debilitating swelling of the foot and lower leg [1–3]. The disease develops when microparticles of aluminum silicates and volcanic silica pass through the skin of people walking barefoot in areas with irritating volcanic clay soil and/or soil covered with red clay, especially in poor farming communities where people do not regularly wash their feet with soap and water [1–5].

There are, however, several risk factors involved in the development of this disease, including family history (supporting a genetic predisposition to the disease), the absence or inadequate use of footwear and poor foot hygiene [1–5]. The disease affects people of all ages and both sexes, but women and girls are disproportionately affected [1].

The disease persists among the poorest populations in the regions of Africa (8.1%), Central America and India (0.21%). Approximately 4 million people are affected in 17 countries worldwide, including 12 in Africa, 3 in Latin America and 2 in Asia [1]. It's worth noting that the disease burden is highest in tropical Africa: around 1.5 million people are affected in Ethiopia, 40,000 in Cameroon, 9,000 in Kenya and 7,000 in Rwanda [1].

In the Democratic Republic of Congo (DRC), epidemiological data remain poorly known, despite sporadic cases already reported [6,7]. However, there are isolated rural communities in the country, particularly those dependent on subsistence farming and those who expose themselves by frequenting mining sites for economic reasons [1,6]. These are all risk factors for the disease, when combined with the absence of footwear or the wearing of unsuitable footwear. Nevertheless, it should be noted that a person exposed to these risk factors does not necessarily develop the disease, but this could be possible through a combination of environmental factors

and genetic predisposition [1–5]. Carrying out surveys throughout the country will enable us to better understand the epidemiology of this non-filarial lymphedema with regard to environmental risk factors and genetic predispositions, which is necessary to reduce avoidable mortality, disability and handicap by providing quality healthcare services to affected patients.

In this case report, we describe the clinical presentation of a patient who initially consulted us for laboratory confirmation of *Mycobacterium ulcerans* infection before a definitive diagnosis was made.

II. Observation

A 55-year-old female patient who has been living for more than 10 years in the mining territory of Kahemba in the province of Kwango in the DRC, and working in the mines for socio-economic purposes. She always lived in an unpaved house with a hard red clay beaten earth floor, and used to stay sometimes stay barefoot at home and also in the mines during her activities. The onset of the condition was marked by the appearance of burning followed by itching and pain in the feet and right leg, around the years 2012-2013. About two to four weeks later, a swelling appeared on part of the foot, which then spread to all the feet and the right leg. Several years later, this swelling spread to both pelvic limbs.

Being in the bush and given the absence of a health care establishment on site, she took medicines of an unknown nature, as well as medicinal plants (for local use and to be consumed orally) which were offered to her, without any improvement. She reported difficulties in getting to work, and avoided people's company because of teasing. The presence of foul-smelling oozing infections prompted her to move to the village for medical care at a suitable health facility. She was treated with antibiotics such as lincomycin, amoxicillin, ampicillin, etc at adult doses parenterally for several days, combined with local treatment of oozing wounds. On physical examination, she was a non-diabetic patient with normal blood pressure (120/80mmHg), and extremely hypertrophied and hyperkeratotic left feet and legs with hard edema and areas of lesions resembling skin fungus (unlabeled). In addition, she had thickened skin and papillomatous growths on the sides of the feet and heels. Swelling of the lower limbs was bilateral and asymmetric, limited to the area just below the knees. There were also nodules and macerations between the toes. Capillary refill was normal in both pelvic limbs, but peripheral pulses could not be detected due to induration and edema, as seen in the two images below ([Figure](#)).



Figure: Presentation of the clinical lesions observed. a) lymphedema of the legs; and b) hypertrophied and hyperkeratotic left leg and foot with outgrowth. ©Photos from personal source.

In addition to routine laboratory tests within normal limits, diagnostic tests for microfilariae and *Mycobacterium ulcerans* carried out at the Parasitology-Mycobacterium Laboratory of the National Institute of Biomedical Research (INRB) were negative. These tests were based on previously established and accepted recommendations, including blood tests under appropriate conditions [8]. In the absence of a reference diagnostic tool, the diagnosis of podoconiosis was based on the clinical picture after exclusion of other causes of lymphedema such as filarial lymphedema, and on the patient's history. The patient was transferred to the university clinics in Kinshasa for appropriate medical care.

III. Discussion

Podoconiosis, also called “dust in the feet”, is considered a stigmatizing public health problem in endemic countries, with an estimated global burden of 4 million people in 32 countries, mainly in tropical countries of Africa, Central and South America, and Southeast Asia [1–5]. The term podoconiosis has been proposed to refer to the non-filarial form of elephantiasis, a non-communicable disease, which can be eradicated by avoiding exposure to irritating volcanic soils through good foot hygiene and the use of socks and shoes [1–5].

The main challenge in combating this disease is the lack of information about the existence of the disease and its differentiation from lymphatic filariasis and other causes of lymphedema in endemic countries, which requires rigorous prevention and control strategies that include avoiding contact with irritating soils (primary prevention) and managing lymphedema morbidity. The disease takes longer to be detected than it should due to the lack of accurate diagnostic tools [1]. Thus, early diagnosis would allow appropriate management and stop the progression of the disease to debilitating stages [1].

Here, we report a case of podoconiosis diagnosed in Kinshasa, Democratic Republic of Congo (DRC), with the aim of drawing the attention of health care providers and policy makers to be attentive to this preventable disease of non-filarial lymphedema in the DRC. Although this is not the first description of the disease in the DRC [6,7], this clinical report sufficiently demonstrates that underreporting of podoconiosis is likely due to difficulties in diagnosis and a low index of suspicion within clinicians. According to a statement by the World Health Organization (WHO), the disease affects people of all ages and both sexes, although women and girls are disproportionately affected [1].

However, the observation made during a meta-analysis by Bikorimana et al. in Rwandan regions reported that female patient had 2 times more likely to contract podoconiosis than male patient; also demonstrated that the disease was significantly prevalent in the 60-year-old age group [5]. This corroborates our observation in a 55-year-old female patient. As described in previous reports, the patient's long-term illness has serious social and economic consequences [1,4,6]. The case described here is that of a mother who worked to support her family in a context of extremely limited resources. Following her illness, she is no longer able to do so or to take care of herself and all those in her care. This observation is therefore consistent with WHO statements showing that 45% of affected patients lose their time of economically productive activity due to the morbidities related to this disease [1]. In Ethiopia, podoconiosis causes the loss of 172,073 disability-adjusted life years each year [1,9].

Several authors argue that the diagnosis of this disease is clinical, depending on whether the patient is in a geographic region at risk, after excluding other causes of lymphedema in a context of hereditary and hygienic risk factors (absence or insufficient wearing of shoes and poor foot hygiene) [10–12]. Other studies have shown that there are factors that reduce the risk of developing this disease, such as belonging to a middle-income household and having access to a certain level of education. Other authors have also shown that podoconiosis only develops in certain subgroups of populations at risk, not in all [11].

An Ethiopian study revealed the presence of familial aggregation, with a high heritability level of 63% [11,12]. In addition, a genomic approach of 194 cases and 203 controls, as well as familial testing, revealed an association between HLA class II variants and podoconiosis, suggesting that it is a T-cell mediated inflammatory disease [11,12].

Treatment is most effective when the disease is diagnosed early as shown by the WHO statement, specifying that the early stages are reversible provided that good foot hygiene and the use of protective footwear are observed. Conversely, the later stages cause considerable economic and social difficulties and, despite adequate treatment, may never completely resolve [1,10–13]. WHO specifies that the disease can be treated with lymphatic decompression therapy even at an advanced stage, either with manual or pneumatic devices, and compression stockings, or with specialized boots [1]. However, reduction surgery is an option for very severe hyperplastic and verrucous elephantiasis [1–3,10–13].

In addition, an advanced state of fibrosis can be found in some patients [1,6,9]. This observation is consistent with our case and we believe that achieving total remission of the lower limbs and feet will not be total. It is therefore essential to take measures to control this disease, such as raising awareness of the population about cleanliness and monitoring those affected, without forgetting the lack of understanding of the environmental trigger to draw attention to the structural inequalities that contribute to the persistence of this avoidable situation [1–5].

The patient's history includes chronic exposure of bare feet to irritating red clay soil and also characteristic elements arguing in favor of podoconiosis. This observation is consistent with those of other investigations carried out elsewhere [2–4,9]. Thus, raising awareness about cleanliness is currently the best way to avoid contamination by this disease.

IV. Conclusion

Podoconiosis diagnosis is often delayed in our limited-resource contexts due to the lack of information on the disease by health care providers and decision-makers, and due to the lack of standardization of diagnostic

tools and protocols. In a risky environmental and hereditary context and after exclusion of other potential causes of lymphedema, the diagnosis of podoconiosis remains possible. More than ten years after the onset of disease manifestations in the patient studied, she was diagnosed on the basis of this set of elements.

Disease awareness campaigns and nationwide epidemiological studies are needed to better understand the burden of this disease with significant socio-economic repercussions.

Conflicts of interest

The authors declare no conflicts of interest.

Authors contributions

MJBK and HNTS: designed the study. MJBK and BZB: drafted the manuscript first version. MJBK and TMI: performed biological analyses. JJTM: supervised the study. All authors reviewed and approved the latest version of the manuscript.

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