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Case Report

Lymphedema due to Podoconiosis in Brazil

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Abstract: Podoconiosis is a neglected tropical disease that causes a type of progressive, non-infectious tropical lymphedema. Aim: The aim is reported one case of the one woman that presented always having problems with allergies and "lumps" appeared on the hallux of the right foot; it resembled a callus, which never disappeared and increased in size. The physical exam revealed several hardened nodular lesions in both feet involving the soles. The patient was submitted to intensive treatment for lymphedema using the Godoy Method® for the lower limbs for five days. The nodules were hardened and constant from the beginning. However, intensive treatment led to a reduction in the volume of the lesions, enabling better plantar flexion and extension as well as a gain in ambulation. However, the characteristics of the evolution of the condition, symptoms and the occurrence of nodules on velvety skin with a mossy kept appearance because are characteristics of podoconiosis. Podoconiosis may have other causes beyond those reported in literature, likely related to the exposure of the limb and associated immunological abnormalities.

Keywords: Podoconiosis; Lymphedema; Diagnosis; Treatment.

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1. Introduction

Podoconiosis is a neglected tropical disease that causes a type of progressive, non-infectious tropical lymphedema mainly affecting the lower part of the leg. This condition results from the long-term exposure of bare feet to soil with red clay derived from volcanic rock and is associated with both poor communities and inadequate foot hygiene [1]. Mineral particles in the soil penetrate the skin and are removed by macrophages of the lymphatic system, causing inflammation and fibrosis of the lumen of the vessel, which leads to the blocking of lymphatic drainage [2, 3].

The lymphatic vessels become gradually obstructed, causing progressive lymphedema with corresponding skin changes in the lower limb. The main differential diagnosis in endemic regions is filariasis. Skin manifestations include dermal nodules and a rough, velvety appearance of the skin, known as "mossy foot", which are pathognomonic of the disease [4]. The main and most severe complication of podoconiosis is acute adenolymphangitis, which presents as extremity warmness and pain, accompanied by fever [2]. Podoconiosis is defined as bilateral but asymmetrical edema, which first develops in the foot and is often confined to the lower part of the leg [5].

Podoconiosis is characterized by a prodromal phase, which involves itchy skin in the forefoot and a burning sensation in the foot and leg. Initial signs may be edema of the forefoot, plantar edema with lymphatic secretion, an increase in marks on the skin, hyperkeratosis with the formation of papilloma resembling moss and stiff toes. The edema is

initially soft and then becomes fibrotic and is often associated with several hard skin nodules. Episodes of acute adenolymphangitis occur, in which the limb becomes warm and painful. These episodes seem to be related to the progression of hard, fibrotic skin. Currently, podoconiosis is only diagnosed clinically based on characteristic signs of the disease and the exclusion of infectious and hereditary causes of lymphedema [6-8].

Podoconiosis is found in more than ten countries of tropical Africa, Central and South America and northeastern India. In Ethiopia, the prevalence of podoconiosis is around 5% in areas with irritating soil [9, 10]. An estimated four million people live with podoconiosis throughout the world [11]. In Brazil, a case was reported as a probable case of podoconiosis, which makes this condition rare in the country [12]. The objective of the study is to report a rare case of podoconiosis in Brazil.

2. Case Report

A 48-year-old woman reported always having problems with allergies and "lumps" would appear in the armpit or inguinal region whenever she hurt her arms or legs, but never with fever. Seven years earlier, a "lump" appeared on the hallux of the right foot; it resembled a callus, which never disappeared and increased in size. Soon afterward, she began to have itching and redness in the region of both ankles, followed by the heels. The sole of the feet began to swell, and it became painful to walk, as if she were "walking on thorns". Multiple nodules began to appear, initially on the right foot and then on the left, approximately 4 cm above the ankle, which worsened over the years. The nodules were soft when they first appeared but became hard after a few days, involving the entire foot (lateral parts and sole). The skin had a mossy characteristic if not washed constantly.

Over the years, the patient became more limited and the limited range of motion of the ankle hindered gait. The patient underwent lymphoscintigraphy, which confirmed the bilateral lymphedema, with an important limitation regarding the ascension of the radiotracer to the inguinal region and with distal dermal reflux. The physical exam revealed several hardened nodular lesions in both feet involving the soles (Figures 1A to 1D). To walk, the patient needed to drag her feet along the ground. The skin had a region with a mossy appearance that, according to the patient, required constant cleaning in order not to worsen.

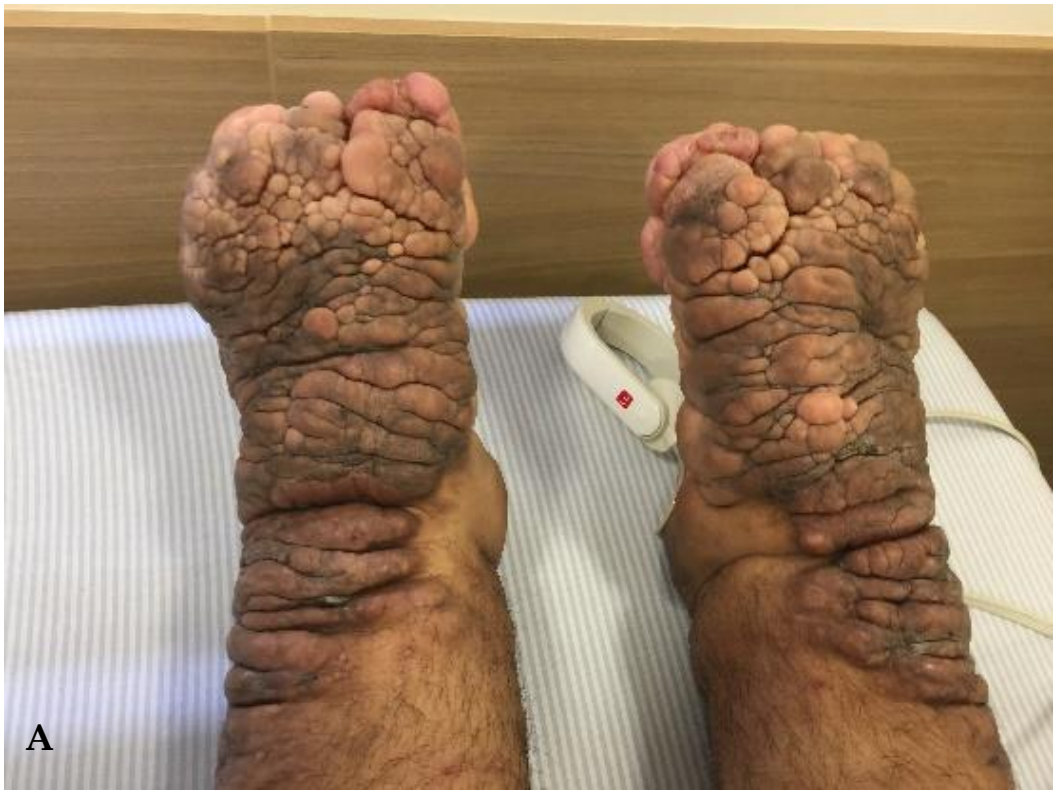
The main differential diagnosis is other causes of primary lymphedema and secondary causes, but none of them have the same characteristics as those of the present study. With more than 10,000 patients with lymphedema treated by the author, no case of primary or secondary lymphedema due to cancer, trauma, or infection has this characteristic in the history and physical examination. The patient states that she has never lived or had contact with regions where there are reports of filariasis in Brazil.

The patient was submitted to intensive treatment for lymphedema using the Godoy Method® for the lower limbs for five days. Treatment consisted of eight hours per day of mechanical lymphatic therapy using an electromechanical device that performs passive plantar flexion and extension movements 28 times per minute; 15 minutes/day of cervical lymphatic therapy, which consists of gentle stretching of the skin of approximately 0.5 cm in the cervical region; approximately one hour/day of manual lymphatic therapy (Godoy Method®); and compression mechanisms (laced stockings made of grosgrain [non-elastic] fabric adapted and adjusted to the size of the limbs). With treatment, the volume of the right leg reduced from 6330 to 5993mL and the volume of the left leg reduced from 6226 to 5945mL in one week. The indication of the Godoy® method is due to its innovation and the only method described proposes the normalization or near normalization of lymphedema [13-18].

Joint mobility improved and the patient was able to walk better. She was instructed to continue treatment at home using the electromechanical device (RAGodoy®) and grosgrain stockings. After one year of treatment, the patient is currently able to walk well and even run and reports a considerable improvement in her quality of life. It has been more than four years since treatment and the patient remains stable, however the patient only

underwent treatment for one week and to achieve normalization or near normalization she needs more weeks of treatment (around four).

Figure 1. A to D show multiple nodules right and left foot.



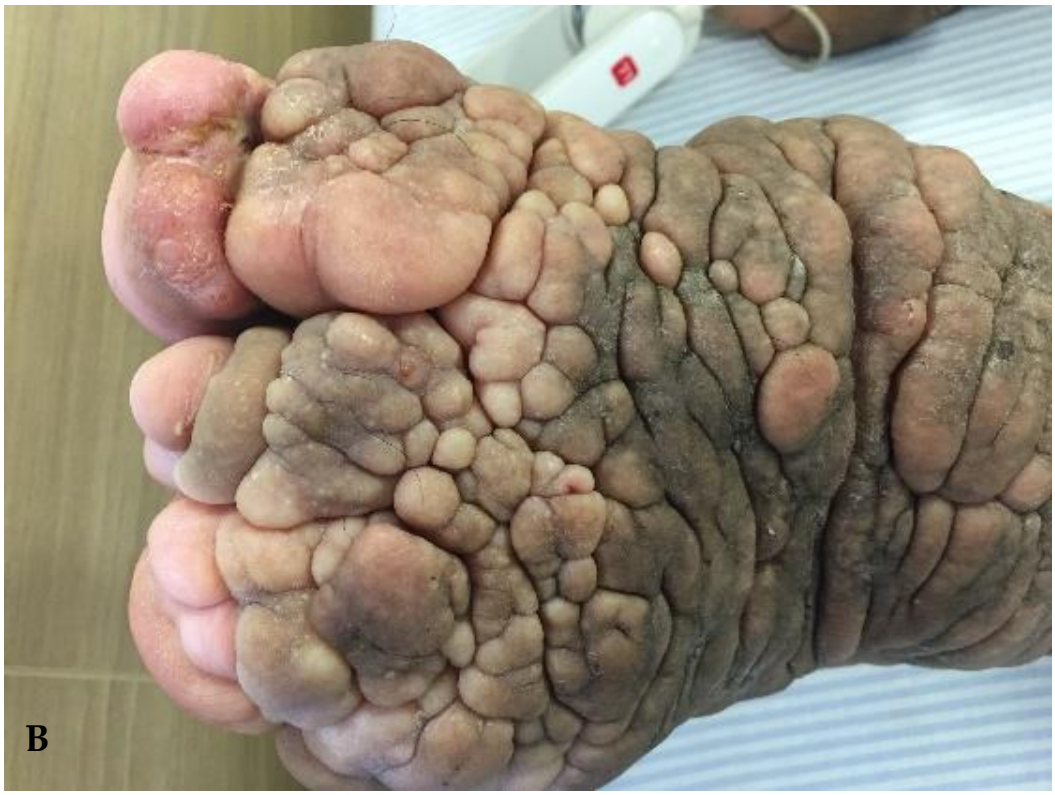
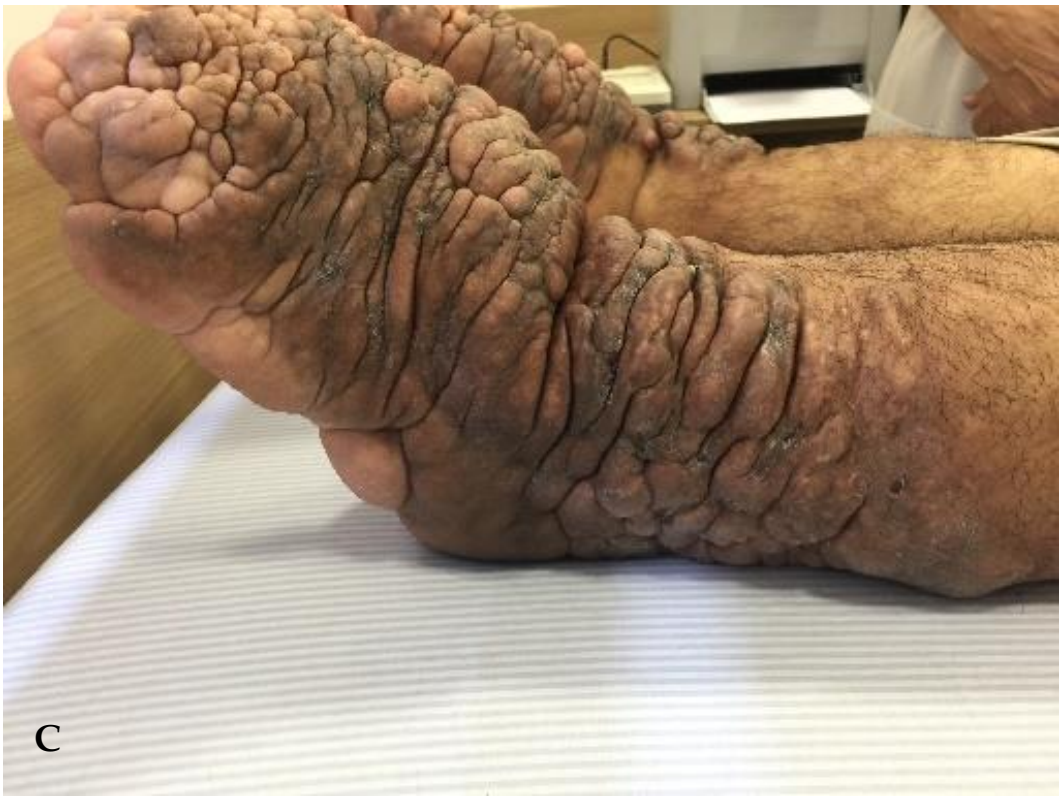


Figure 1. A to D show multiple nodules right and left foot (*continued*).



3. Discussion and conclusion

The present study reports the first case in the Brazilian literature of podoconiosis in a 55-year-old patient. The diagnosis of this condition is based on the patient history and physical examination. There is no routine complementary exam for such cases. In regions where podoconiosis is common, the differential diagnosis is performed with filariasis.

The history and clinical evolution reveal typical characteristics of podoconiosis and skin with a mossy appearance is pathognomonic for the disease. However, the patient in the present case did not have a history of walking barefoot over red clay soil derived from volcanic rock and no other cases of the disease occurred in the region where she lived. One of the main hypotheses is that mineral particles in the soil penetrate the skin and are

removed by macrophages of the lymphatic system, causing inflammation and fibrosis of the lumen of the vessel. Hygiene is another aspect. This patient had a higher education and constant care of her feet.

The region marked by nodules was limited to approximately 4 cm above the ankles since the onset of signs and symptoms. The nodules were hardened and constant from the beginning. However, intensive treatment led to a reduction in the volume of the lesions, enabling better plantar flexion and extension as well as a gain in ambulation. In endemic regions, hygienic care, wearing shoes and the use of compression therapy improves the clinical condition of these patients. In Brazil, the differential diagnosis includes the investigation of primary and secondary lymphedema. However, the characteristics of the evolution of the condition, symptoms and the occurrence of nodules on velvety skin with a mossy appearance are characteristics of podoconiosis.

Approximately one year has passed since the beginning of treatment and the patient is maintaining the clinical improvements achieved with intensive treatment through the maintenance of care at home. No evident complications have occurred in this period and the improvement of the limbs has been constant. The goal is to accelerate the reduction in fibrosis through another week of intensive treatment, which is the aim of the method [12-14]. The intensive Godoy Method® proposes the normalization or near normalization of fibrosis, leg and arm [12, 13], and keep the results [15], which is currently being proven by biopsies taken before and after treatment [16-19]. Therefore, this is the goal for the patient, but it is fundamental to eliminate the causal factors of the lesions.

The physio pathological hypothesis is that this patient has some immunological defects by which micro traumas, such as walking barefoot at home, can trigger the condition. She had no history of travelling to regions with red clay soil or of having walked barefoot in such regions. The bilateral nature and simultaneous onset suggest a triggering factor of the physio pathological process. Podoconiosis may have other causes beyond those reported in the literature, likely related to the exposure of the limb and associated immunological abnormalities.

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Research Ethics Committee Approval: This study received approval from the institutional review board (IRB) of the São Jose do Rio Preto School of Medicine-FAMERP #4.903.458. The term consent for published and included image was signed by patient. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration.

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Conflicts of Interest: The authors declare no conflicts of interest.

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