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Case Report

Elephantiasis nostras verrucosa: case report

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ABSTRACT

Lymphedema is a clinical condition in which an accumulation of macromolecules occurs in the interstitial space and an atypical evolution known as elephantiasis nostras verrucosa may occur. The objective of this study is to report on the clinical evolution and analysis of the progression of a 56-year-old patient with a diagnosis of primary bilateral stage II lymphoedema. With an atypical presentation of it, identified as elephantiasis nostra verrucosa. The patient had suffered from lymphoestatic edema since he was 10 years old, having previously received ineffective treatments until he arrived at the Clinica Godoy in Sao Jose de Rio Preto Brazil, where he underwent an intensive therapy procedure treatment model proposed by Godoy and Godoy; in which favorable results were obtained in a short period of time. Lymphedema is a chronic disease that is manageable but not curable. It is also difficult to be treated, especially if it has progressed to a late stage such as elephantiasis nostras verrucosa associated the continuous and repetitive infectious and inflammatory processes over time must have contributed to the evolution of the fibrotic pattern. Intensive treatment based on the Godoy method® is possible reduce in short period of time the volume and improve fibrosis and quality of life this patient.

Keywords: Lymphedema, Treatment, Elephantiasis, Nostras verrucosa

INTRODUCTION

Lymphedema is a clinical condition in which an accumulation of macromolecules occurs in the interstitial space derived from an insufficiency of the lymphatic system that results in fluid retention in a segment of the body and formation of edema. It has primary and secondary causes where in the primaries the person is born with some type of alteration in the lymphatic system. In the secondary lymphatics, they are born with a normal limb and during the course of life some injuries such as surgery, radiotherapy, erysipelas and others can give rise to lymphedema.[1]

An atypical evolution of lymphedema is observed in the literature known as elephantiasis nostra. Repetitive or chronic infectious processes constitute the main hypothesis for this form of clinical evolution.[2,3]

There is no single form of treatment for lymphedema, association of therapies is usually recommended. Manual and mechanical lymphatic drainage, lymphomyokinetic activities and exercises, compression garments and mechanisms are part of physical therapy, but social and psychological aspects could be considered.[4,5]

The objective of this study is to report on the clinical evolution and analysis of the progression of a 56-year-old

patient diagnosed with stage II primary bilateral lymphedema. With an atypical presentation of it, identified as elephantiasis nostra verrucosa.

The patient had suffered from lymphostatic edema since he was 10 years old, having received previous ineffective treatments until he arrived at the Clínica Godoy in Sao Jose de Rio Preto, Brazil, where he underwent an intensive care process according to the treatment model proposed by Godoy and Godoy; in which favorable results were obtained in a short period of time.

CASE REPORT

We describe the case, male, of a 56-year-old patient who began to present edema in the right foot at the age of 10, followed by several episodes of infection and continuous progression of the skin lesions. At the age of 15, she began to present edema in the left foot, also followed by several episodes of infection, but less than in the right. At 20 years of age, she had an episode of erysipelas that led to skin necrosis in the anterior region of the distal third of the right leg. In this episode, a skin graft was performed that evolved with hypertrophic healing of the wound. In these infectious episodes, she used antibiotics, since the only name she remembers is benzetacil. This medication was also used as a prophylaxis for erysipelas, but he still had other infectious outbreaks. She comments that she underwent treatment with the use of pneumatic compression, but she could not tell for how long and denies having undergone lymphatic drainage interventions. Previously, compression devices such as stockings and bandages were not used.

In the physical examination, intense fibrosis was detected in the feet and fingers with constant discharge. Nodular lesions affecting the foot and fingers are observed on the right foot, as shown in Figure 1a and b. The nails of the right foot were practically destroyed, as shown in the Figure. A diagnosis of elephantiasis nostras verrucosa was made and an intensive treatment based on the Godoy method® was proposed. Before and after the treatment, bioimpedance was performed, which showed a significant reduction in edema in both limbs. In the right leg the volume (ml) detected by bioimpedance with the Inbody S 10 device was 10.23 ml, which was later reduced to 8.07 ml and in the left leg it went from 9.30 ml to 6.71 litres. Mechanical lymphatic drainage with RAGodoy® was used, which passively performs plantar flexion and extension around 25 to 30 cycles per minute, associated with cervical lymphatic therapy that consists of about 30 soft stimuli in the cervical region per minute for 15 minutes per day, as well as elastic bandages of medium elasticity and manual lymphatic therapy, which is a manual technique used to accelerate the reduction of edema.[5-7] A handmade inelastic compression garment was made of grosgrain fabric.[8] Throughout the treatment there was a reduction of important edema, as well as reduction of the secretion and fibrosis of the affected limb. The study was approved ethical committee Faculdade de Medicina de Sao Jose do Rio Preto#2.336.784. The examination of the patient was conducted according to the Declaration of Helsinki principles. The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient signed consent term.

Figure 1 (a and b): Nodular lesions affecting the foot and toes.

DISCUSSION

The present study shows the evolution of a patient with congenital early primary lymphedema that evolved towards elephantiasis de nostra. This is an unusual form of presentation of lymphedema in these patients and few descriptions are found in the literature.[2] The literature suggests episodes of sudden infection as the cause of this evolution.[3] We consider it important to report and document these cases, due to the limitation and the little information or availability of the same in the current scientific literature.

In this study, what is striking is the chronicity of the process without the classic evolution towards a significantly increased pattern of edema. What is observed is the large amount of skin fibrosis and loss of skin elasticity and the constant secretion that, according to his clinical history, gave rise to constant episodes of active or chronic infection. Another fact that draws attention is the nails that are practically destroyed, probably due to infectious pictures. Thus, the continuous and repetitive infectious and inflammatory processes over time must have contributed to the evolution of the fibrotic pattern.

It is also important to mention how the gradual loss of skin elasticity resulted in a significant reduction in joint mobility capacity in the most hardened regions, such as the ankle and metacarpophalangeal joints. The interdigital regions presented chronic and suppurating wounds that did

not heal spontaneously. It is possible to attribute this behavior to the chronicity of the edema, the fibrotic consistency of the skin, and the underlying chronic inflammatory process as a result of chronic infection. Another point to take into account and on which it is worth paying attention is how the skin evolved to give rise to the formation of nodular lesions in the distal region of his foot and toes, having as background the characteristics of the edema and behavior the skin.

CONCLUSION

In conclusion, lymphedema is a chronic disease that is manageable but not curable. It is also difficult to be treated, especially if it has progressed to a late stage such as elephantiasis nostras verrucosa associated the continuous and repetitive infectious and inflammatory processes over time must have contributed to the evolution of the fibrotic pattern. Intensive treatment based on the Godoy method® is possible reduce in short period of time the volume and improve fibrosis and quality of life this patient.

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