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Ainhum. Presentation of Clinical and Sonographic Findings in A Colombian Patient

Sandra Muvdi*
Claudia González**

*Dermatologist, MsC Clinical Epidemiology, Instituto Dermatológico Federico Lleras. Private Practice Clínica Marly

**Radiologist, MSK and Dermatological Ultrasound, Instituto Diagnóstico Médico. IDIME.Bogotá. Colombia, Address: AUTOPISTA NORTE 122-68, Email: claud_gonzalezdiaz@yahoo.com

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SUMMARY

Introduction: Ainhum or Spontaneous Dactylosis (SD) is an infrequent dermatological disease characterized by the development of a constricting band, generally on the fifth toe that can end in self-mutilation.

Objective: To present the clinical and imaging findings (radiography and high-resolution ultrasound) of a 77-year-old Colombian patient with Ainhum.

Conclusion: Although the diagnosis of Ainhum is clinical, the use of high-resolution ultrasound can confirm the presence of the characteristic constricting band of the disease and to be considered as a useful tool to establish the staging and prognosis of the entity together with conventional radiography.

KEYWORDS

Ainhum, Spontaneous dactylosis, High resolution ultrasound, Pseudoainhum, Amputation

INTRODUCTION

The word Ainhum means fissure in the Iago language (Brazil) or saber cut in the Yoruba language (Nigeria) [1]. It is an idiopathic disease, relatively rare, which has been described mainly in black people. It consists of a spontaneous dactylosis [2] that leads to the self-mutilation of one or more fingers, generally although not exclusively, of the fifth toes. Its etiology is unknown, although factors such as chronic trauma from walking barefoot has been associated with its physiopathogenesis [3]. We present a case of a Colombian black patient, with an emphasis on ultrasound findings, which have not previously reported in the medical literature, which allow establishing an early diagnosis of the disease and proper management of it [4].

CLINICAL CASE

This is a 77-year-old black male patient from Bagadó (Chocó, Colombia) who reports a one-year history of a sensation of mass and edema of the base of the fifth left toe, with burning sensation and pain; who has empirically treated with magnesium sulfate baths, with partial improvement of pain and with reappearance of symptoms for periods of two to three months. As important antecedents, he refers to having worked as a fisherman and walking barefoot in the jungle for long periods of time. He has received treatment with antidepressants and does not mention other important antecedents.

Physical examination of the left toe shows a circular constricting band with a fissure at the base of the toe and a protrusion of the phalanges (Figures 1). On the right toe, there is a bulbous protrusion of the soft tissue base without a fissure (Figure 2). Distal pulses were present, although weak, vascular evaluation and the Doppler study showed non-occlusive arteriosclerotic disease and ruled out that the clinical findings

were of vascular origin. Additionally, there was no hyperkeratosis. Clinically, it was not clear if the phalangeal protrusion corresponded to exostosis or soft tissue injury, so a high-resolution soft tissue ultrasound was requested. Ultrasonography shows a well-defined, hyperechoic, linear structure delimited by a markedly hypoechoic band with well-defined edges corresponding to the constrictor band, which covers the three layers of the skin without bone involvement. (Figures 3 & 4) No other findings such as soft tissue mass or exostosis were found. Radiography of the left 5 toe showed slight rotation of the proximal phalanx without bone resorption or other alterations (Figure 5). The clinical and imaging findings establish the diagnosis of Ainhum.



Figure 1: Clinical image of the left five toe shows a circular constricting band with a fissure at the base of the toe and a protrusion of the phalanx



Figure 2: Bulbous protrusion of the soft tissue base without fissure on the right fifth toe

DISCUSSION

The first case of spontaneous dactylosis was reported in the world literature in the State of Bahia in Brazil in 1867 by da Silva Lima who used the term Ainhum [5] to describe a form of spontaneous dactylosis, which affected the fifth toe of a “Quilombola”. The word ainhum means fissure in the language of the Nagos tribe of Brazil and ayum of Nigeria which means saw.

Regarding the epidemiology of Ainhum, it is observed that it mainly affects black people from Africa, Asia, North America and Brazil [6]. However, there are few cases of SD in white individuals published in the literature [7]. Its prevalence has been reported in 2.2% in Nigeria, 0.2% in Congo and 0.015% in Panama [8,9]. In Colombia there are previous isolated reports of the entity [10,11]. Men are more affected than women, in a 2: 1 ratio, generally between 30 to 50 years [12] and there may often be a family history of the disease [13].

The etiological factor of spontaneous dactylosis is unknown, however, the repetitive trauma of walking barefoot in the jungle is described as a trigger for it [14]. The patient in our case is black and has a clear history of walking barefoot for long periods of time as a fisherman. Other associated factors described are: mycosis, skin infections, syphilis, scleroderma, peripheral neurological problems, plant toxins, osteomyelitis, and schistosomiasis [15-17].

SD usually involves the toes, generally, but not exclusively, the fifth toes, unilaterally or bilaterally [18,19]. There are few reported cases of finger involvement [20].

The pathognomonic clinical finding, corresponds to the presence of a constricting band that compromises the base of the toes that conditions a fissure with bulges of the distal soft tissues, eversion, reabsorption and with the auto-amputation progression of the compromised toe [21]. The entity is painful and can often become overinfected.

The diagnosis is clinical, since as the constrictor band is described, it is characteristic of the disease. The imaging test generally practiced is conventional radiography, which allows establishing the presence or not of osteolysis of the involved phalanges and establishing their classification and management. In the longest series published in the literature, Daccarett describes the radiological findings that are classified into four phases [22].

Stage I, there is no bone involvement and in the soft tissues the radio-lucid constricting ring or band can be observed at the base of the phalanx.

Stage II, the constrictor ring progresses generating an “hourglass” configuration of the soft tissues and the phalanx presents external rotation giving the appearance of the lateral projection in the anteroposterior projection of the finger.

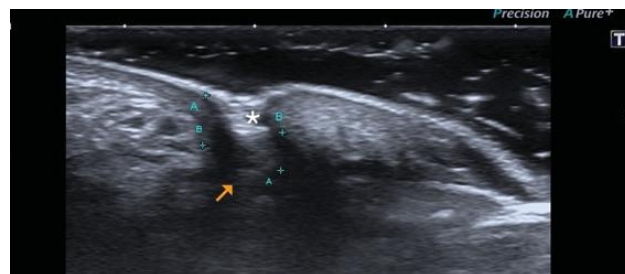


Figure 3: High-resolution longitudinal ultrasound image with an 18 MHz linear transducer, demonstrates a well-defined, hyperechoic (white asterisk) linear structure delimited by a markedly hypoechoic band with well-defined edges corresponding to the constricting band, (yellow arrow) covering the three layers of skin without bone compromise. Between calipers A + the length of the band is demonstrated, calipers B + show the thickness of the band

Stage III the presence of osteolysis is characteristic and is described as the “hanging finger” image, bone resorption begins in the middle aspect of the phalanx and progresses laterally, the narrowest part of the bone may appear on osteoporotic radiography or less frequently sclerotic.

Stage IV: spontaneous amputation of the hanging finger occurs, the bone portion of the residual stump is triangular or “Iceberg” in this stage clinically at the time of amputation there may be increased pain and bleeding in the layer, in addition an ulcer may develop at the base of the amputation. If there is an infection, periostitis can also be observed on the radiograph [23]. Our patient presented a stage II with the rotation of the phalanx without osteolysis in the fifth toe of the left foot (Figure 5). In the fifth toe of the right foot, the radiograph was considered normal.

In the literature, there are no previous published reports of ultrasound findings, as found in our patient. Ultrasound shows the development of a linear band, markedly hypoechoic with well-defined edges, corresponding to the constricting band, which involves soft tissues with variable depth from the epidermis to the deep plane, depending on the stage of the disease. The hypo-echogenicity of the band on ultrasound is interpreted as secondary to the presence of fibrotic tissue in the constrictor ring that has been observed in the histological reports of the Ainhum [24]. In the middle third of the band there is a hyperechoic line that correlates with the presence of air inside the fissure. The ultrasound to be performed for the characterization of SD is high-resolution ultrasound, following the guidelines recommended by expert groups on the subject in dermatological ultrasound [25,26]. The use of a high resolution linear transducer (18 to 24 MHz) allows to accurately identify the exact length of the constricting band in millimeters, its thickness and the accuracy of the soft tissue layer that it compromises, providing relevant information since with radiography, only the bone involvement and the presence or absence of the constricting radiolucid ring can be established or not, but not its real extension in the soft tissue layers. In previous published reports, ultrasound has been used for patients with suspected Ainhum, which demonstrated the presence of a foreign body in soft tissues, corresponding to a false Ainhum [27].

The differential diagnosis should be made with other entities that involve the development of a constrictive fibrotic band in the phalanges, such as Mibelli’s porokeratosis, erythropoietic protoporphyria, scleroderma, psoriasis, neuropathic plaque, leprosy, syphilis, Raynaud’s disease, diabetes mellitus and syringomyelia [28-32].

Regarding the treatment of the disease in the early stages I-II, the resection of the constricting band followed by Z-plasty, can alleviate pain and decrease the risk of progression to self-amputation. In stages III-IV, surgical imputation may be recommended [33]. In our patient, when he was in stage I-II, surgical management with Z-plasty was recommended.

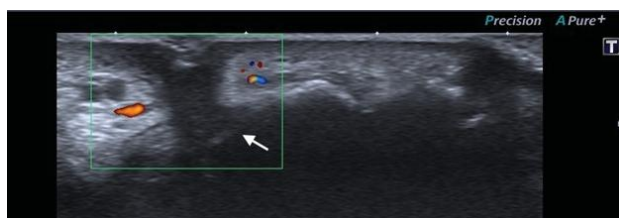


Figure 4: Longitudinal high-resolution ultrasound image with an 18 MHz linear transducer and color Doppler analysis, confirms the marked hypo echogenicity of the constricting band without flow on Doppler examination, observing the peripheral arterial vessels with flow inside (White arrow)

CONCLUSION

SD is a rare entity, with variable prevalence. We present the clinical and imaging findings of a Colombian patient with SD. The patient worked as a fisherman, with a history of walking barefoot for long periods, which, as described in the discussion, has been associated with the development of the entity. The use of conventional radiography for the diagnosis and staging of the entity is well known. However, there are no previous published reports of Ainhum ultrasound findings. The high-resolution ultrasound performed on the patient confirmed the presence of the constricting band and the ultrasound aspect of the band is described. As observed, ultrasound can identify the constricting band, its size, characteristics and accurately identify the layer or layers of compromised tissue, providing useful and accurate information for the diagnosis, prognosis and treatment of the entity.



Figure 5: AP radiograph of the left five toe shows slight rotation of the proximal phalanx without bone resorption or other alterations

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