

Ainhum (Or Dactylolysis Spontaneous) at the Fousseyni Daou Hospital in Kayes about a Case

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Abstract

Ainhum or spontaneous dactylolysis is a progressive constriction of the digito-plantar fold of the fifth toe which leads after several years of evolution to a spontaneous amputation of the toe. Classically, a distinction is made between “true Ainhum” of unknown cause, which affects only blacks and those of African (sub-Saharan) descent; “pseudo-Ainhum”, which follows various causes such as an inflammatory flange or foreign body constriction; and finally, palmo-plantar keratoderma, of genetic origin, such as Vohwinkel disease. We report a case of Ainhum received at stage III of the pathology and who benefited from an amputation of the fifth toe.

Keywords

Ainhum, Spontaneous Dactylolysis, Idiopathic, Amputation

1. Introduction

Etymologically, the word “Ainhum” is derived from the Nago word (Brazil) meaning “fissure” or the Yoruba word (Nigeria) meaning “to saw or cut”. Ainhum is a relatively rare idiopathic dermatosis [1] [2]. The first case reported in Brazil was by Silva Lima in 1867 [3]. The prevalence varies from 0.015% to 2% of the population in African countries and few reports in white individuals are found in the literature [4]. The disease is characterized by the appearance of a

progressive ring of constriction usually on the fifth toe, which can lead to spontaneous self-amputation of the affected toe [5] [6]. A distinction must be made between “true Ainhum”, which is of unknown cause and affects only blacks and those of African (sub-Saharan) descent [5] [7]; “pseudo-Ainhum” [2] [8] [9], which follows a variety of causes such as an inflammatory flange or foreign body constriction; and finally, palmo-plantar keratoderma ainhumoides, of genetic origin, such as Vohwinkel’s disease [10]. This is a typically African dermatological pathology that can occur at any age and in both sexes, which is why we initiated this study (in collaboration with the dermatovenerology department) in order to establish a principle of management of this pathology. An early diagnosis and management can avoid mutilating deformations as well as amputations which are generally sources of psychological after-effects.

2. Observation

This was a 66 year old female patient with a low socioeconomic level, admitted to the hospital on November 6, 2021 with a 15 month history of a painful circular constriction band at the base of her right fifth toe without any notion of trauma, ligature or ring on the toe and without any previous consultation. The main reason for her consultation was the increase in the intensity of the pain, which was becoming more and more insomniac. There was no previous ulceration on the base of the toe. There was no history of diabetes, ischemic heart disease, peripheral vascular disease, skin pathology, HIV/AIDS, or psychiatric illness. She also had no history of surgery and was not aware of any family history of Ainhum. She was not an alcoholic or smoker. There was a notion of barefoot walking.

On examination, the patient was in good general condition with clear consciousness and was afebrile. Her blood pressure was 146/82 mmHg, pulse was 105 beats/minute and regular, respiration was 16 cycles per minute and temperature was 37.4°C.

Examination of the right foot showed stricture of the base of the toe by a keratosis band with the presence of a traditional black powder that the patient used for self-care (**Figure 1**). The toe was oedematous, cyanotic and painful. There was diffuse plantar hyperkeratosis (**Figure 2**). The examination of the left foot was normal. Biological examinations (blood count: Leukocytes at $4.5 \times 10^3/\text{ml}$, Fasting blood glucose was 1.02 g/dl; creatinine was 83 mmol/ml Group O Rhesus+) were normal. The foot X-ray noted images of osteolysis of the middle phalanx at the level of the stricture zone. We performed a local anesthesia of the fifth toe, an incision at the base of the toe followed by its dissection and then an amputation taking away both phalanges (**Figure 3** and **Figure 4**). X-rays of the right foot showed osteolysis of the middle phalanx (**Figure 5**).

An analgesic treatment based on paracetamol 1000 mg every 06 hours; an antibiotic prophylaxis by 2 G of Amoxicillin + Clavulanic acid and a dressing. The postoperative course was simple.



Figure 1. Keratotic band on the 5th toe.



Figure 2. Diffuse plantar hyperkeratosis.



Figure 3. Amputation under local anesthesia of the 5th toe.



Figure 4. Amputation under local anesthesia of the 5th toe.



Figure 5. Osteolysis of the middle phalanx of the 5th toe.

3. Discussion

Ainhum disease occurs mainly in Black Africa; it is of unknown etiology, is characterized by the appearance of stricture around the fifth toe, and is seen mainly in black-skinned adults in tropical environments [1]. Neumann used the term Pseudo-Ainhum to refer to other forms of constriction of the fingers and toes [7]. It is an extremely rare dermatologic condition with reported prevalence rates of 2.2% [1], 0.2% [8], and 0.015% [9] in Nigeria, Congo, and Panama, respectively. In their study, Marcos *et al.* observed during 22 years (1977 to 1999), on radiographs of 6000 suspected patients, 102 cases (1.7%) presented spontaneous dactylolysis. All patients were black Africans and in this series, dactylolysis was not associated with any other pathology [10].

During twelve years, it was the 5th case encountered in our hospital, *i.e.* a prevalence of 0.011%. There is a preponderance of the disease in people aged between 20 and 50 years [5]. Men are more prone than women with a male/female sex ratio of 2:1 [5]. Classically, the disease is most often bilateral and affects the fifth toe in 75% of cases [11]. It should be noted that a few cases of isolated involvement of the finger [8] and the big toe [12] have been reported. Generally, patients have a family history of Ainhum [13]. Our patient, although black African with unilateral involvement, was not aware of any case of Ainhum in her family.

Ainhum is commonly reported as an idiopathic pathology [5]. Nevertheless, some authors have put forward some etiological hypotheses such as infections (mycotic, mycobacterial), trauma, decreased vascular supply, peripheral neuropathies and genetic pathologies (keratodermas) [14]. Trauma-related cases are often trivial and may go unnoticed, such as walking barefoot in the tropics. In our case, apart from barefoot walking, none of the above-mentioned causes were mentioned in our patient. This supports the hypothesis of an idiopathic etiology as previously described in the literature [5] [7].

The diagnosis of Ainhum is essentially clinical, described as a spot diagnosis by some authors [15]. The pathogenesis involves the development of a flexion groove of the phalanx, with progressive complete circumferential constriction of the phalanx [5]. The natural history evolves into spontaneous self-amputation [5].

Although considered an ad hoc diagnosis, the diagnostic criteria established for Ainhum involves three conditions: 1) soft tissue constriction with bulbous enlargement of the toe, 2) phalangeal bone thinning, and 3) phalangeal lysis [1], all of which were observed in the present case.

It is worth mentioning that there are four clinical stages of Ainhum [1]: 1) a clavus develops, which evolves into an annular fissure around the toe, 2) the toe becomes globular downstream of the groove, associated with bone resorption and arterial narrowing, 3) very painful bone becomes detached at the joint with hyper mobility of the toe, 4) an auto bloodless amputation of the toe with severe pain. Our patient presented with stage 4 and we performed surgical amputation of the toe to ensure proper wound healing.

Browne SG [9] and Morand L [13] reported that Ainhum was associated with plantar hyperkeratosis, implying that this association could be closely related to the etiology of Ainhum. Our patient illustrated an idiopathic etiology as described by several authors [16] [17].

On radiographs, the lesions include a radiolucent ring, constricting the base of the toe, bone resorption, and osteolysis mostly of the middle and distal phalanges with a characteristic taper [11]. The X-ray of our patient's right foot showed osteolysis of the middle phalanx of the fifth toe.

The differential diagnosis arises with pseudo-aenhums which are most often secondary to: congenital pathologies, such as Streeter's dysplasia [10] or a mutilating keratoderma hereditarium (Vohwinkel's syndrome) [10], constriction re-

sulting from trauma or related to other diseases such as scleroderma, syphilis, leprosy, atypical keratoderma diabetes yaws or vascular gangrene [15] [18]. In our case, we could not rule out these differential diagnoses due to infrastructural limitations.

In the early stages of the disease, non-operative treatment using topical or injectable salicylate preparations [14], corticosteroids [19], or retinoids [5] is recommended. Surgical management of stage 1 and 2 ainhumas involves plasty to release the constrictive base of the toe [1] [14] [20]. Surgical amputation is the mainstay of treatment for stages 3 and 4 [5], this was in our patient. Amputation helps to relieve pain and prevent surgical site infections [13]. If left untreated, self-amputation, secondary infections, and locomotor imbalance may complicate ainhum [5].

4. Conclusion

Ainhum is a rare mutilating dermatological condition and is more frequent in men from sub-Saharan Africa. The authors emphasize the need for a high index of suspicion on the part of health care providers, especially those in wound care, for prompt diagnosis and management to prevent self-amputation, permanent deformity and disability, psychological trauma, and surgical site infections.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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