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Ainhum and Syndactyly in the Foot: A Rare Association in an Adult

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Abstract **Case Report**

Ainhum is a rare pathology, mainly observed in the black population. After several years of evolution, it leads to spontaneous amputation of a toe. Congenital syndactyly is a malformation of the hands or feet. however, to our knowledge, the association between ainhum and congenital syndactyly in an adult is unknown in the literature. We report the first case of double bilateral aihum associated with congenital syndactyly in a black adult in Niger treated by surgical method for ainhum and therapeutic abstention for syndactylies with a good evolution. The association between ainhum and foot syndactyly is extremely rare and remains to be elucidated by other studies.

Keywords: Ainhum, syndactyly, congenital, adult, Niger.

INTRODUCTION

Ainhum, or spontaneous dactylosis, is a condition of unknown etiology, mainly affecting black population [1]. The condition begins as a hyperkeratotic fissure at the base of the toe, then develops as a constructive fibrous band around the toe, usually leading to spontaneous amputation a few years later (around 5 years).

Congenital syndactyly is common in the feet. The most frequent, autosomal dominant, familial involvement concerns the second commissure. Syndactyly may be simple (soft tissue fusion) or complex (bony fusion) [2]. In some cases, they may be associated with other malformations [2]. However, the association between ainhum and syndactyly remains poorly understood in the literature. To our knowledge, no cases have been reported to date in the literature about this association.

We report the first case of an association between ainhum and congenital syndactyly in the foot bilaterally in an adult in Niger.

CASE REPORT

This was a 64-year-old farmer with a history of self-amputation of the fifth toe of the right foot dating back some 15 years. He presented with stabbing pain on walking and ulcerative striation of the fifth toe of the left foot. The onset of symptoms was thought to date back some three years, with progressive worsening of the lesion in a non-infectious, non-traumatic context. The patient's general condition was good.

Clinical examination revealed a striated circular ulceration on the fifth toe of the left foot, and an incidental finding of syndactyly between the second and third toes on both feet (Figure a). Examination of the right foot revealed a healed amputation stump (self-amputation) of the fifth toe of the right foot, dating back some 15 years, and syndactyly of the second and third radii (Figure a).

Vascular and nerve examinations were normal. The rest of the clinical examination and biological tests were normal.

X-rays of both forefeet showed osteolysis at the fifth radius, with no bone fusion between the bones of the second and the third radii (Figure b, c).

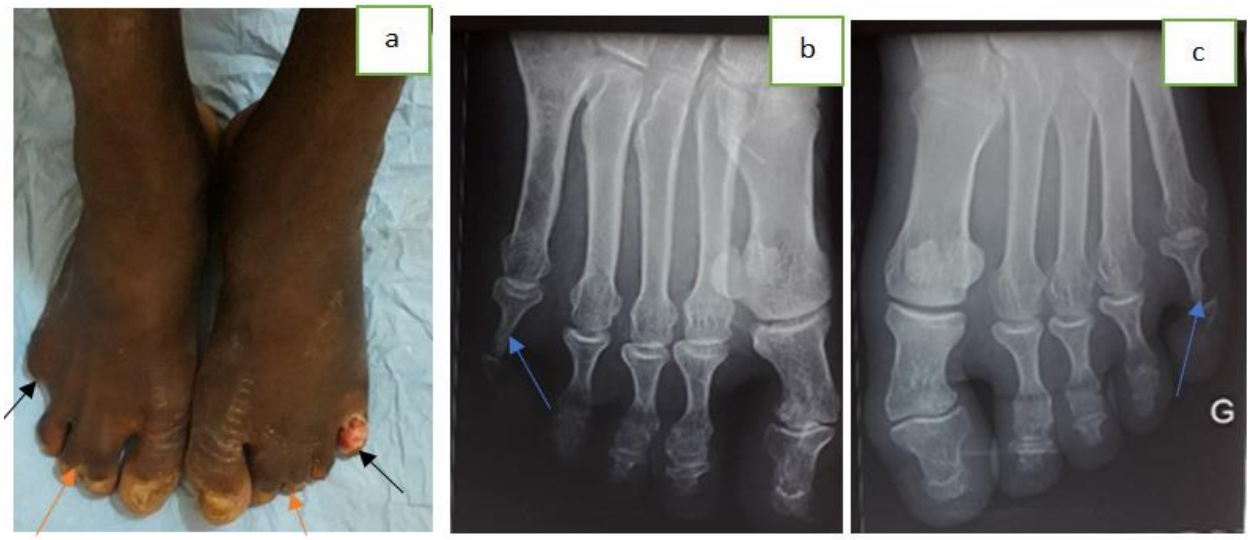

Figure: (a) Local aspect of the right foot with healed self-amputation stump and left foot with circular ulceration at the base of the fifth toe and bilateral syndactyly of second and third radii of both feet. (b) and (c) radiographs of right and left forefoot showing successively complete osteolysis of the third phalanges, head, body, and base of intermediate phalanges of the fifth radii on left and right

- Black arrows: striated circular ulceration on the fifth toe of the left foot and healed amputation stump (self-amputation) of the fifth toe of the right foot.
- Orange arrows: syndactyly between the second and third toes on both feet.
- Blue arrows: osteolysis of the first and second phalanx of the 5th toe on the right and left corresponding to the seat of the ainhum.

The diagnosis of a double association of bilateral ainhum stage 2 according to COLE's classification on the left fifth ray and stage 4 on the right fifth ray (scar) as well as bilateral syndactyly of the second and the third ray of both feet was then made.

Surgical management for the left fifth toe was performed. With local anesthesia, we performed an amputation at the base of the left fifth toe associate with a cover plasty of the amputation stump. Concerning syndactyly, therapeutic abstention was observed.

The evolution was marked by the good healing of the amputation stump and the patient resumed walking normally without pain.

DISCUSSION

Ainhum or spontaneous dactylolysis is a rare pathology whose etiopathogenesis remains poorly elucidated. Indeed, several authors in the literature agree that this pathology is mainly observed in dark-skinned people from Africa, America, or the West Indies, regardless of gender [3]. It can occur at any age, but more frequently in adult subjects between 21 and 59 years of age, with a slow clinical course ranging from 18 months to 10 years [4, 5].

Syndactylies are congenital malformations due to a differentiation disorder of the mesoblast between the sixth and eighth weeks. They affect both hands and feet, with variable functional repercussions depending on the affected radius [2, 6, 7]. They may occur alone or in association with other malformations [2, 8].

However, no association between ainhum and congenital syndactyly has been reported in the literature, particularly in the black race.

The case reported in our study specifically evokes the evolving clinical picture of ainhum of the fifth toe bilaterally, in its most described form, evolving over more than years, culminating in spontaneous amputation on the right, with a particular association with bilateral congenital syndactyly of the feet.

According to Cole [9], the natural evolution of ainhum involves four degrees of increasing severity:

- Stage 1 corresponds to the groove. This is the constriction of soft tissue on the tibial surface of the fifth toe,
- Grade 2 is ulceration of the bottom of the groove,
- Grade 3 reflects bone involvement. Osteolysis, with a tapered appearance of the bone, is opposite the proximal interphalangeal joint. The head of the proximal phalanx is misaligned,
- Degree 4 corresponds to self-amputation.

Bone damage is constant in ainhum (figures b, c). It depends on the complexity of the syndactyly picture [2, 7].

Concerning syndactylies, therapeutic abstention was observed for our patient. In fact, it is reported in the literature that for simple syndactyly from

the second commissure in the foot, most teams recommend therapeutic abstention. There are no functional consequences [2].

In short, this case report raises the question of ainhum, a disease whose etiopathogenesis is still poorly understood, and of a unknown clinical association between ainhum and congenital syndactyly of the foot which deserves to be elucidated.

Finally, ainhum must be distinguished from pseudo-ainhum. The latter is defined as any case of self-amputation not associated with the classic spontaneous ainhum observed in Africans, the etiology of which is unknown [10].

CONCLUSION

Ainhum is a rare pathology, most often found in the black population, with a very slow evolution. The association of this pathology, mainly in the black race, with congenital syndactyly of the foot remains poorly understood. This study reports the first case of association between the two pathologies in a black man and raises the question of the etiopathogenesis of a disease that remains unresolved to this day, and of a rare malformative association in an adult whose link remains to be investigated.

Conflict of Interest: The authors declare no conflict of interest related to this work.

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