

Eruptive Villous Cysts: An Unusual Complication of Suppurative Keloids, First Case Report

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Introduction

Keloids are pathological scars secondary to aberrant collagen proliferation and impaired apoptosis of myofibroblasts involved in the healing process [1]. These occur more frequently in Afro-descendants and Hispanic populations (4.5-16%), presenting in all cases a chronic and recurrent course, affecting the quality of life secondary to aesthetic compromise [1,2].

They are benign fibroproliferative tumors of the reticular dermis secondary to trauma, which generate an exaggerated growth scar and affect the adjacent skin, associated with pain or pruritus in 40% of cases, and which do not resolve spontaneously [3]. Among its complications, is the appearance of cystic lesions that generate inflammation, erythema, pain, and discharge, known as suppurative keloids (25-26%), whose pathophysiological mechanism results from follicular fibrous entrapment in the scar [4,5], has been described. Another abnormal healing process is the eruptive hairy cysts, which are the result of infundibular occlusion that leads to the retention of hairy fragments accompanied by keratin, which then forms drainage fistulous tracts [6].

The evaluation with high-resolution color Doppler ultrasound allows the evaluation of the anatomical characteristics and the

evolution of the different dermatological lesions, including keloids and hairy cysts [7,8].

This non-invasive imaging diagnostic tool is the one that best allows the identification and characterization of these lesions, establishing their precise diagnosis when performed under dermatological ultrasound guidance [9].

Presentation of the case

A 35-year-old male patient with a history of cystic acne conglobata, with numerous keloid scars on the anterior chest (Figure 1A). He presented with a two-month-old lesion, located on a keloid scar in the sternal region, associated with pruritus, local irritation, and serohematic secretion, without evidencing an increase in size.

Physical examination revealed in the anterior thorax an extensive, elevated scar-like plaque that exceeded the limits of the initial lesion and a non-fetid serohematic drainage point on the sternal region, with pain on local palpation (Figure 1B). Dermoscopy revealed serohematic crusts, milky red-white areas without structure, scant desquamation, irregular linear vessels, and some comma vessels at the periphery of the lesion (Figure 1C).

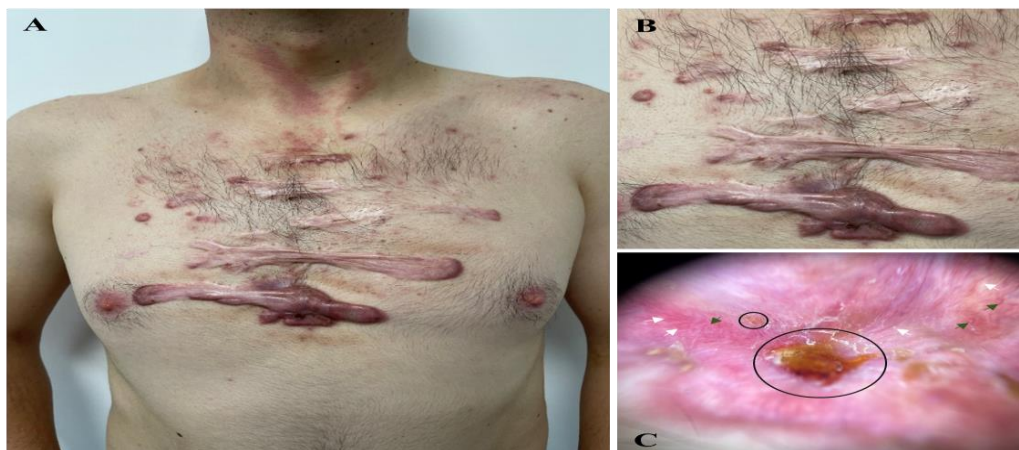


Figure 1: A, Numerous keloid scars on the anterior thorax. B, Extensive plaque with a keloid scar appearance with a sternal serohematic drainage point. C, Dermoscopy: serohematic crusts (circles), milky red-white areas without structure, scant desquamation, irregular linear vessels (white arrows) and some comma vessels (green arrows) at the periphery of the lesion.

The high-resolution soft tissue ultrasound with Doppler analysis reported multiple cysts distributed in the lower area of the lesion and towards the subcutaneous tissue. Dilation of multiple follicles in the presence of at least five cystic structures with remains of keratin fragments inside, suggestive of hairy cysts that fistulate towards the middle third of the keloid, two of them reaching the surface with a clinically evident hole in the epidermis and the rest fistulized inside the keloid (Figure 2).

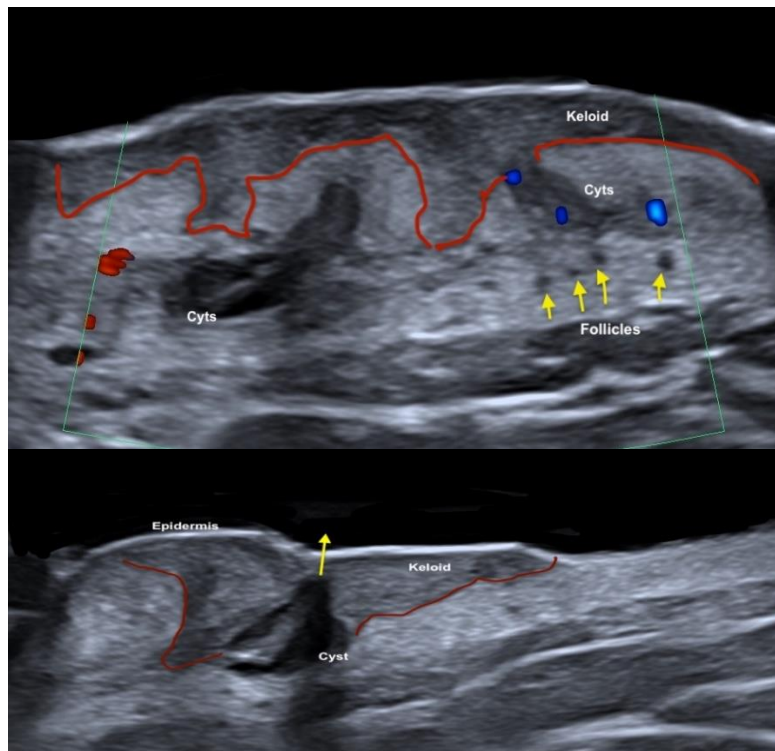


Figure 2: Multiple keloid scars (delimited by a red line) with the presence of follicular dilatations and cystic structures compatible with villous cysts associated with fistulous tracts. The yellow arrow shows how the hairy cyst protrudes from the base of the keloid and heads toward the surface, trying to drain into the epidermis.

Discussion

Suppurative keloids are keloid scars that are complicated by the formation of fistulous tracts, edema, drainage, pain, and in some cases, with systemic symptoms, presenting a higher incidence in African American men, with lesions on the beard, neck, and presternal region [5,10].

Onwukwe, in 1987 described three pathophysiological processes associated with this entity. First, a focus of necrosis is secondary to the increase in the size of the lesion, which exceeds the perfusion capacity and its vasculature. Second, fibrosis in the ostium of the pilosebaceous unit with a phenomenon of liquefaction and drainage through the fistulous tracts. And third, the maceration effect when the keloid appears in special areas such as intertriginous areas. These phenomena together generate a greater risk of superinfection in keloids [10]. Delaleu et al in 2015, described that the most frequent histopathological finding in the pathogenesis of these lesions was the obstruction of the pilosebaceous ostium and stated that the rupture of the follicle favors the foreign body reaction, giving rise to the development of the keloid [4]. It is worth mentioning that a history of acne is described in up to 47% of patients with suppurative keloids [4].

The eruptive villous cysts represent an infrequent benign disorder secondary to infundibular occlusion [11]. They were first described in 1977, by Esterly et al. in two pediatric patients with the appearance of follicular lesions without associated inflammation, located on the extensor surface of the extremities and in the anterior thorax [11,12]. Some authors consider them as hamartomas originating in the matrix of the hair follicle and in the outer sheath of the hair, between the infundibulum and the

isthmus, and others describe them as cysts that block the exit of the terminal hair [13,14].

Its cause is unknown. An autosomal dominant inheritance pattern has been described, and there are cases of sporadic appearance during the first two decades of life, without being associated with specific triggering factors [14,15]. Some cases appear late (after 30 years), after sun exposure, trauma, and pregnancy [16].

Clinically, they present as small papules with a dome-shaped surface, single or multiple, between 1 and 4 mm in diameter and in some cases up to 7 mm, which can be euchromic, yellowish, brown, erythematous, bluish or black, with a keratotic or umbilicate [16]. Eruptive hairy cysts can be soft or firm to the touch and, in some cases, present discharge of white or grayish material, but they are usually asymptomatic, and rarely pruritic [12,16]. They are found mainly on the anterior thorax and face, but they can also appear on the neck, anterior and posterior trunk, armpits, inguinal region, buttocks, and even on the eyelids, sites with a greater number of pilosebaceous units and apocrine glands [14,16].

Zhu et al. described the dermoscopic findings of hairy cysts, including homogeneous yellow circular structures without blood vessels or inflammation, as well as gray-blue lines near the follicular openings [12,17].

Yoo et al. described the presence of arboriform vessels, irregular linear vessels, and comma vessels in the dermoscopy of keloids, reflecting increased vascularity in the tissue [18]. The latter is a consequence of the inflammatory process and angiogenesis

secondary to excessive tissue growth, coinciding with the dermoscopy performed on the patient [19].

Ultrasonography is a useful tool for monitoring keloids, which allows for evaluating characteristics such as depth, echogenicity, and vascularization [19]. Thanks to its high collagen content, altered metabolism, and aberrant dermal network, it increases the water content of the extracellular matrix, which translates into areas of greater hypoechogenicity [20,21]. The classic appearance on ultrasound is a hypoechoic thickening of the dermis that displaces the epidermis upward and follows the long axis of the skin layers [22]. They may present parallel linear hyperechoic bands that correspond to thick collagen fascicles and some pseudo nodular areas. Fistulous tracts have also been described under keloids that can arise secondary to mechanical stress and collagen degeneration, among others [19,22]. Usually, an increase in arterial and venous flow velocity is observed on color Doppler evaluation [19]. The ultrasonographic findings in eruptive villous cysts are nonspecific, being able to find hypoechoic nodules with well-defined edges with a thin capsule in the dermis or subcutaneous cell tissue, and inside they can have linear hyperechoic fragments corresponding to keratin and calcium remains. They can even present a fistulous path or punctum that drains towards the epidermis [23].

The treatment of eruptive hairy cysts is cosmetic. However, some can be self-resolving by transepidermal removal or by the formation of foreign body-type giant cells that react by forming granulomas. Keratolytics, topical or oral retinoids, curettage, cauterization, and CO₂ laser vaporization have been used for removal [13,14]. For keloids, treatment is a challenge due to the risk of recurrence. Despite this, there are surgical and non-surgical interventions that reduce its progression and improve aesthetics and even the function of the affected tissue [1].

Conclusion

The first case found in the reviewed literature described a patient with a history of cystic acne conglobate and a suppurative keloid scar associated with the formation of eruptive hairy cysts on its surface. Since these diseases share a common pathophysiological pathway derived from alterations in the pilosebaceous follicle, their exhaustive clinical and imaging evaluation is of vital importance. Biopsy of a keloid is avoided to reduce its risk of worsening and in this case, high-resolution skin color Doppler ultrasound becomes a fundamental diagnostic tool not only in the evolutionary behavior of this type of lesions but also facilitates its characterization and adequate therapeutic direction.

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