Idiopathic Calcinosis Cutis of the Scrotum: A Case Report

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Calcinosis cutis—calcification in soft tissue—is a rare benign disease that is separated into the following subtypes: dystrophic, iatrogenic, metastatic, calciphylaxis, and idiopathic. One of common site of calcinosis cutis is the scrotum. The nodules slowly grow for years or decades. The characteristic of calcinosis cutis of the scrotum is generally asymptomatic, yellowish marble-like, hard, polypoidal, solitary, or multiple. However, the pathogenesis of this nodule remains ambiguous and controversial. Thus, we reviewed possible causes and therapeutic consideration of calcinosis cutis of the scrotum. (Korean J Urol Oncol 2017;15(2):88-91)

Key Words: Calcinosis • Skin • Scrotum

INTRODUCTION

Various factors can cause scrotal masses to be formed, from benign causes such as spermatocele, epididymitis, orchitis, hematocele, or inguinal hernia, to malignant causes such as testicular cancer. Even skin tumors must be considered as possible diagnoses. Clinicians can presume the diagnosis by the appearance of a tumor, history taking (onset, clinical course, pain, sexual contact history, etc.), physical examination (mobility, tenderness), and laboratory data (especially regarding tumor markers). The most crucial point for diagnosis is histological factors. Idiopathic calcinosis cutis of the scrotum is one diagnosis of a scrotal mass. The feature of calcinosis cutis of the scrotum is generally slowly growing yellowish nodules that consist of calcification with surrounding foreign body-type granulomatous

inflammation. Since Lewinski1 first described calcinosis cutis of the scrotum in 1883, and Shapiro et al.2 established idiopathic scrotal calcinosis as a distinct entity in 1970, several cases had been reported worldwide. However, most of the etiology and pathophysiology remains unknown and controversial. Thus, we present a case of a 35-year-old male patient with massive idiopathic scrotal calcinosis.

CASE REPORT

A 35-year-old male presented for evaluation with multiple scrotal nodules that had developed over the past 10 years. These nodules had gradually increased in number and size over the years. He did not complain of any symptoms such as itching or pain. His complaint was that these scrotal lesions were bizarre, and that he therefore felt shame for showing the scrotal lesions to his son. He denied any history of scrotal trauma, past medical history, or family history. On physical examination, the whole area of the scrotum lesion measured 10 cm×8 cm. Each nodule was yellowish-white and was 0.2–2 cm large. The arrangement was irregular and had definite boundaries (Fig. 1A). Upon scrotal ultrasonography, no specific lesion was found in
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Fig. 1. (A) Clinical photograph of the scrotum showing multiple small skin-colored papules. (B) Surgical excision and primary closure were performed.

Fig. 2. No specific lesion in the testes or epididymis on scrotal ultrasonography. Nodular echogenic lesions with posterior shadowing just below were shown. Color Doppler ultrasonography found no vascularity.

The testes or epididymis. Nodular echogenic lesions with posterior shadowing just below were shown. No vascularity was found in the nodules upon color Doppler ultrasonography (Fig. 2). Regarding laboratory data including lipid profile, the blood chemistry such as the serum calcium, phosphorus, and parathyroid hormone levels stayed within normal range.

Although the nodules’ features were nonspecific, we assessed the lesion as epidermal cysts or xanthomata and commenced surgical intervention. Under local anesthesia, the excisional biopsy of one nodule was done without any complication and the patient was discharged just after the operation. After a week, histologic examination showed intradermal calcification, which led us to suspect calcinosis cutis. Upon examination under a microscope, the scrotal nodule showed amorphous deposits of basophilic staining calcium crystal in the dermis and calcium deposits were confirmed on Von Kossa and alizarin red stains. Moreover, a large deposit of calcium was shown, presenting foreign body reactions such as giant cells, inflammatory infiltration, and fibrosis at a high power field (Fig. 3).

We changed the assessment to idiopathic scrotal calcinosis and scheduled a reoperation to excise the remnant lesion under spinal anesthesia. During the operation, we concerned about skin grafts or flaps. However, even though the skin defect lesions were rather large, primary closure was possible because of redundant and elastic scrotal skin (Fig. 1B). We sutured the wound site using 3-0 vicryl. On postoperative day 2, the patient had no complications and was discharged. After a week, histologic examination showed consistency with calcinosis cutis. The postoperative course was uneventful and the cosmetic results were good. The patient was followed up for 3 years without any skin lesion recurrence.

DISCUSSION

Calcinosis cutis—the deposition of insoluble calcium salts in the soft tissue—is a rare benign disease. This disease is categorized according to etiology: dystrophic, iatrogenic, metastatic, calciphylaxis, and idiopathic. Calcinosis cutis can appear in any skin lesion. One common site of calcinosis cutis is the scrotum.

Calcinosis cutis is a rare benign condition in the scrotal skin,
Fig. 3. (A) Amorphous deposits of basophilic materials are present in the dermis (deep blue stained calcium deposit) (H&E, ×10). (B) A large deposit of calcium was shown presenting foreign body reactions such as giant cells, inflammatory infiltrate, and fibrosis (H&E, ×200).

that featured slowly growing yellowish nodules consisting of calcification with surrounding foreign body-type granulomatous inflammation. Among four types of scrotal calcinosis, the idiopathic type has typical features: the presence of connective tissue disease, tissue injury or the absence of systemic metabolic disorder. In addition, other types of scrotal calcinosis must be pre-excluded.

Shortly after this disorder was first reported by Lewinski in 1883, there was a second validated report by some researchers. After a lot of time, Shapiro et al. suggested the term “idiopathic scrotal calcinosis” as a disease entity in 1970, and many cases have been introduced in the last 30 years worldwide. Dubey et al. reported 100 patients with scrotal calcinosis. The age of the patients was in the range 15–77 years and the mean age was 31.5 years. However, most of the etiology and pathophysiology remain unknown and controversial.

One controversial point is the role of epidermal cysts, which were discussed by Swinehart and Golitz in 3 cases of scrotal calcinosis, and some were calcified with partial or total disruption of the epithelial walls, surrounding by inflammatory cells. Song et al. noted a hypothesis based on histopathology. They suggested that intracystic keratinous material accumulated in the cyst and disruption of the epithelial walls occurred subsequently. This triggers an inflammatory reaction with resorption of the cyst walls and the keratinous material. Eventually, calcific materials in the soft tissue only remained. Meanwhile, King et al. suggested dystrophic calcification of the dartoic muscle to be the primarily shown before calcification in the soft tissue noted, since epidermal cysts must have not be superior to calcification. Furthermore, based on the research of Hicheri et al., no evidence of cystic structure was found around the calcified deposits.

This condition generally appears in adolescence or early adulthood with the normal metabolism in calcium and phosphate. These nodules tend to increase in number and size over time; the nodules are hard and yellowish, and consist of deposits of calcium and phosphates, and are of various sizes in the scale of millimeter to centimeter.

The nodules are asymptomatic in most cases, and in other complicated cases such as infection there are reports of pain and itching. The risk of recurrence is also controversial. Clinically, it may be not definitely distinguished from epidermal cyst, steatocystoma, and cutaneous horn. Also it may be confused with lipoma, fibroma, angiokeratoma, and lymphangioma. Therefore, the diagnosis is usually based on histopathology, featured by calcified materials of various sizes surrounded by an inflammatory changes. Chakrabarti and Sharma framed fine needle aspiration cytology as a diagnostic tool of this rare disease that can help avoid unnecessary surgery.

The main reason for intervention is cosmetic. Other reasons include pruritus and ulceration. The treatment of choice is surgical excision, which gives good clinical results. Because of the laxity of the scrotal skin, practicing multiple nodular excisions
allowed for excellent scrotal coverage and good cosmetic results. The surgical excision may be limited to the skin level, because nodules are generally localized within the dermis. Smaller lesions are amenable to novel pinch punch biopsy, and larger lesions may require wide excision and direct closure. If the lesion involves the whole scrotum or frequent recurrence occurs, extensive scrotal reconstruction may be needed. In our case, there was a massive occurrence of the nodules, but complete excision and primary closure were possible. The rate of recurrence is also controversial, and some authors insist on the frequent recurrence after primary excision. Recurrence may be due to the leftover microscopic calcification foci. Therefore, the surgical approach should be perfect and the extent of excision must include the whole lesion to avoid recurrence.

**CONFLICT OF INTEREST**

The authors claim no conflicts of interest.

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