American Journal of Clinical Case Reports

Case Report

Idiopathic Sacrococcygeal Calcinosis Cutis

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Abstract

Calcinosis cutis is deposition of calcium and phosphorus in the skin. It is a rare benign disease. Idiopathic calcinosis cutis is when there is no any remarkable cause of skin calcification. Although many literatures described commonly affected site by calcinosis cutis is the scrotum, but here we report an uncommon site of calcinosis cutis, which is sacrococcygeal region. A 45 year-old man presented with a ten years history of nodular lesions at sacrococcygeal region. The characteristic of lesion at sacrococcygeal region was painful, hard, brownish and yellowish nodules of different size. Histopathological analysis showed dermal layer of collagen with dense clumps, lumpy salt deposits. However, the pathogenesis and etiology of these nodules remain ambiguous and controversial. After reviewing all possible causes and therapeutic considerations of sacrococcygeal calcinosis cutis, we found it idiopathic.

Keywords: Calcinosis cutis; Sacrococcygeal; Skin calcification

Introduction

Calcinosis cutis is a rare disease defined as the deposition of insoluble calcium salts in cutaneous tissues [1]. The etiology of calcinosis cutis is typically separated into the following categories: dystrophic, metastatic, idiopathic, iatrogenic, or calciphylaxis [2]. The term idiopathic calcification is reserved for skin calcification that is associated with neither an underlying tissue injury nor a systemic disorder [3]. Dystrophic calcinosis cutis is a fairly common condition, whereas idiopathic cases are very rare [1]. When dystrophic calcification occurs locally and consists of only a few deposits of calcium salts, it is referred to as calcinosis circumscripta. When the deposits are large and widespread, it is termed calcinosis universalis [4]. Clinicians can do the diagnosis by history taking, considering the appearance of a tumor, physical examination which focuses on mobility, pain, or tenderness and the results of the laboratory. The most important point for diagnosis is histological factors. Even though the literatures point out the possibility that calcinosis cutis can appear at any region, we could not find any reported case of calcinosis cutis at sacrococcygeal region. We herein report a case of a 45-year-old male patient with an idiopathic sacrococcygeal calcinosis cutis.

Case Presentation

A 45-year-old, apparently healthy man, presented for evaluation with sacrococcygeal paining mass that had developed over ten years. In the beginning, it was small as a size of coin and it increased in two years later. Before, the patient did not complain of symptoms such as itching or pain. His complaint was that these sacrococcygeal lesions were bizarre, and that he therefore felt shame for showing the sacrococcygeal lesions to anybody else. After eight years, he started to experience pain and itch but which were tolerable. The patient stated that he had never experienced any sacrococcygeal disease like trauma, inflammatory symptoms or infection. He denied any history of sacrococcygeal trauma, past medical history, or family history. Physical examination revealed the whole sacrococcygeal region with a mass measured 6 cm × 5 cm and so many hard brownish and yellowish nodules of different size: 0.2 cm – 2 cm large. The arrangement was irregular and had definite boundaries. Laboratory analysis including lipid profile, the blood chemistry such as the serum calcium and phosphorus found within normal range. Tissue biopsy of the lesion has been done. Histological results showed intradermal calcifications, which led us to suspect calcinosis cutis (Figure 1). Histological section stained with Haematoxylineosin ofsacrococcygeal nodule showed dermal layer of collagen with dense clumps, lumpy salt deposits (Figure 2). Moreover, a large deposit of calcium was shown, presenting hyperkeratosis, acanthosis cell layer thickening. The diagnosis of idiopathic sacrococcygeal calcinosis cutis was then confirmed. We decided surgical excision as treatment.

Discussion

Calcinosis cutis is characterized by the deposition of insoluble calcium salts in the skin and subcutaneous tissue. Calcinosis cutis can appear in any skin region. One common site of calcinosis cutis is thescrotum [5] but here we report a case of uncommon place of sacrococcygeal region. Dystrophic calcinosis cutis is the most common subtype and is often seen in connective tissue disease, in certain inherited disorders, in cutaneous neoplasms, and as a result of trauma [3]. This doesn't concern our case because there was no history of sacrococcygeal trauma in this case. Metastatic calcinosis

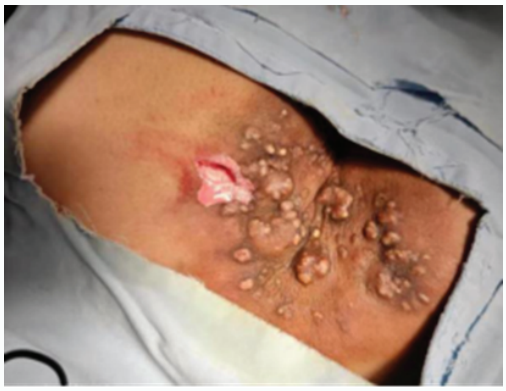

Figure 1: Skin biopsy of sacroccygeal lesion show intradermal calcifications.

Citation: Ntawuyamara E, Xiaoxuan S, Cai D. Idiopathic Sacrococcygeal Calcinosis Cutis. Am J Clin Case Rep. 2020;1(1):1010.

Publisher Name: Medtext Publications LLC

Manuscript compiled: Oct 16th, 2020

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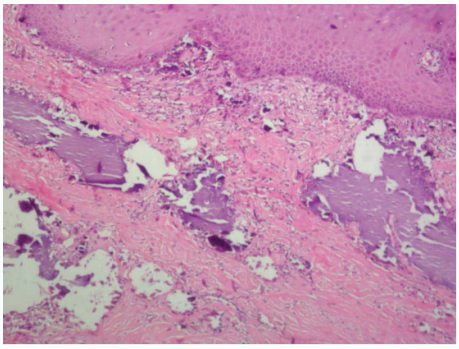

Figure 2: Histopathology of a biopsy specimen of sacrococcygeal mass shows dermal layer of collagen with dense clumps, lumpy salt deposits (HE stain; original magnification ×40).

cutis occurs due to abnormal calcium or phosphate metabolism [3]. In this case, the patient had normal calcium and phosphate test results. Iatrogenic calcinosis cutis occurs as a result of medical causes, such as extravasation of intravenous calcium gluconate therapy [6] but here no medical act documented. Calciphylaxis occurs in the context of end-stage renal disease when there is deposition of calcium in blood vessels in the dermis or subcutaneous fat, and it presents with different clinical skin findings [3]. The patient had no renal disease at all. Among five types of calcinosis cutis, the idiopathichas typical features: it occurs in the absence of known tissue injury or systemic metabolic defect [3]. It has been proved in this case where all laboratory examinations were normal even the serum calcium. The main clinical manifestations are hard papules, nodules or lumps. A white chalky and grit-like substance is discharged after ulceration. The deposited calcium salts are mainly amorphous calcium phosphate, a small amount of calcium carbonate and very little apatite. The exact mechanism of skin calcium is still unclear and it is generally believed that this disease is caused by the release of calcium ions from mitochondria of the damaged muscle cells which promotes mineralization [7]. Saad et al. [8] believe that idiopathic skin calcification is formed by calcification after inflammation of epithelial cysts. There are various treatment options of calcinosis cutis including warfarin, bisphosphonates, minocycline, ceftriaxone, diltiazem, aluminium hydroxide, probenecid, intralesional corticosteroids, intravenous immunoglobulin, curettage, surgical excision, carbon dioxide laser, and extracorporeal shock wave lithotripsy [9]. In this case we preferred surgical excision as it has been shown to be the treatment of choice in idiopathic calcinosis cutis.

Conclusion

Idiopathic sacrococcygeal calcinosis cutis must be distinguished from warts, xanthomatosis, and osteoma of the skin, gout nodules, and progressive bone hyperplasia and calcified fibrous tumors. It is important to differentiate idiopathic calcinosis cutis with other subtypes of calcinosis cutis. Idiopathic calcinosis cutis can also appear in other areas such as the sacrococcygeal described in this case. With the theories of pathogenesis of idiopathic calcinosis cutis, this case report call for further studies.

Funding Sources

2015QLMS16

References

1. A Guermazi A, Grigoryan M, Cordoliani F, Kérob D. Unusually diffuse idiopathic calcinosis cutis. Clin Rheumatol. 2007;26(2):268-70.
2. J Abbott, EH Kowalski, S Klein, R DeShazo, CM Hull. Iatrogenic calcinosis cutis secondary to calcium chloride successfully treated with topical sodium thiosulfate. JAAD Case Rep. 2020;6(3):181-3.
3. N Reiter, L El-Shabrawi, B Leinweber, A Berghold, E Aberer. Calcinosis cutis: Part I. Diagnostic pathway. J Am Acad Dermatol. 2011;65(1):1-12.
4. JS Walsh, JA Fairley. Calcifying disorders of the skin. J Am Acad Dermatol. 1995;33(5):693-706.
5. MMA Syed, A Rajbhandari, U Paudel. Idiopathic calcinosis cutis of the scrotum: A case report and review of the literature. J Med Case Rep. 2018;12(1):88-91.
6. D Goldminz, R Barnhill, J McGuire, KS Stenn. Calcinosis Cutis Following Extravasation of Calcium Chloride. Arch Dermatol. 1988;124(6):922-5.
7. M Kavala. An extremely severe case of cutaneous calcinosis complicating adult dermatomyositis. Clin Exp Dermatol. 2009;34(1):115-6.
8. AG Saad, GS Zaatari. Scrotal calcinosis: is it idiopathic?. Urology. 2001;57(2):365.
9. N Reiter, L El-Shabrawi, B Leinweber, A Berghold, E Aberer. Calcinosis cutis: Part II. Treatment options. J Am Acad Dermatol. 2011;65(1):15-22.