

Idiopathic calcinosis cutis causing cubital tunnel syndrome: A case report and review of literature

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Abstract

Calcinosis cutis is a type of heterotopic calcification where abnormal calcium deposition occurs in skin or subcutaneous tissue. Among the subtypes of calcinosis cutis, the idiopathic variety occurs without underlying biochemical calcium abnormality. We report a rare case of idiopathic calcinosis cutis causing cubital tunnel syndrome. A 63-year-old female presented with pain and numbness in the ulnar aspect of her left hand. The X-ray of the left elbow showed deposition of radiopaque material on the posteromedial aspect. Her nerve conduction study showed evidence of ulnar nerve compression at the elbow supporting the diagnosis of ulnar nerve compression by the mass of calcium deposition. Surgical exploration was performed, and significant ulnar nerve compression was noted due to the mass effect of the calcium deposition. Excision of the mass and ulnar nerve decompression with anterior transposition was performed with satisfactory outcomes. Although calcinosis cutis causing cubital tunnel syndrome has been previously reported, all patients had some form of calcium dysregulation. We report the first case of ulnar nerve compression at the cubital tunnel due to idiopathic calcinosis cutis. Excision of the mass and ulnar nerve decompression with anterior transposition was successful in our patient despite the incomplete excision of the calcium deposition.

Keywords

Calcinosis cutis, idiopathic calcinosis cutis, cubital tunnel syndrome, ulnar nerve entrapment

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Introduction

Calcinosis cutis (CC) is a type of heterotopic calcification where abnormal calcium deposition occurs in skin or subcutaneous tissue.¹ Among the subtypes of CC, the idiopathic variety occurs without underlying biochemical calcium abnormality or primary local tissue injury predisposing to calcium deposition. Apart from clinical features like pain, nodules, ulceration, and calcium extrusion, peripheral nerve compressions are rare.¹ We report a rare case of idiopathic CC causing cubital tunnel syndrome.

Case presentation

A 63-year-old female presented with pain and numbness in the ulnar aspect of her left hand associated with a progressively enlarging swelling over the medial aspect of the elbow for 5 months durations. She had a history of painful swellings of several fingertips with whitish nodules, and

surgical excision confirmed the diagnosis of CC. She did not have any symptoms to suggest arthritis or other connective tissue disorders. Her biochemical investigations had normal serum calcium and phosphate levels. All other organ function tests including renal functions were normal. On clinical examination, she had paresthesia of her ulnar aspect of the left hand with swelling over the medial aspect of the elbow and an associated Tinel sign. Patient did not have any evidence of muscle atrophy or weakness. The

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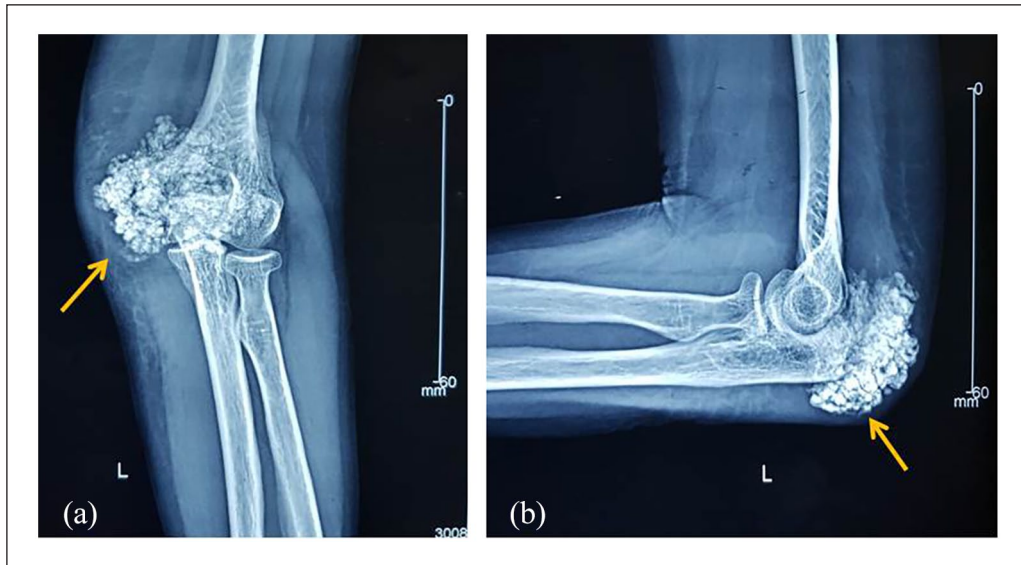


Figure 1. X-ray of the left elbow. (a) Antero posterior. (b) Lateral views. Orange arrow showing the deposition of radiopaque material.

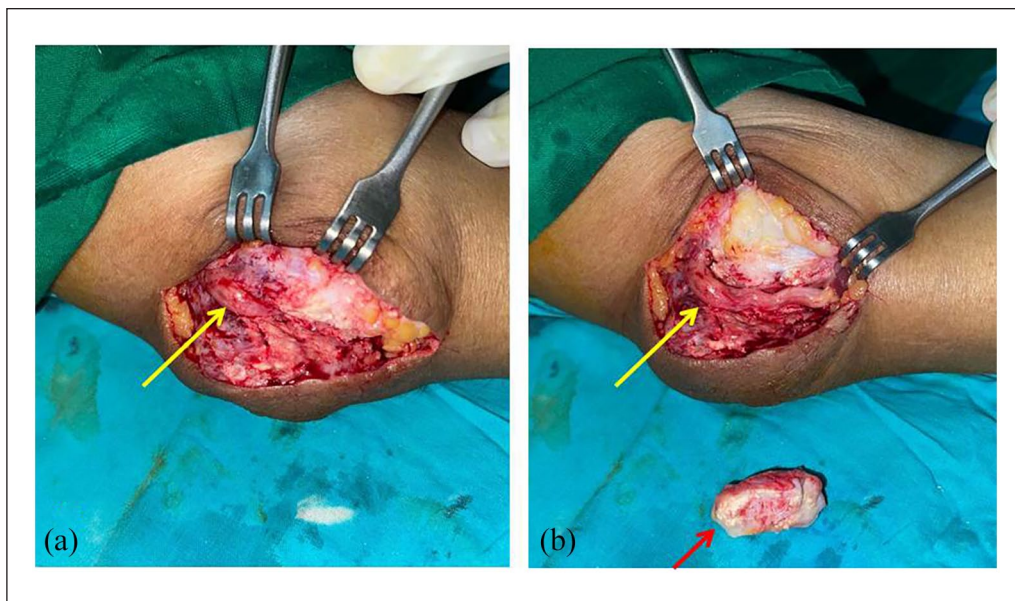


Figure 2. Intraoperative picture of the medial aspect of the left elbow. (a) Entrapped ulnar nerve (yellow arrow) inside the mass. (b) Decompressed ulnar nerve (yellow arrow) and excised mass (red arrow).

X-ray of the left elbow showed deposition of radiopaque material on the posteromedial aspect (Figure 1). Although useful, further imaging such as computed tomography or magnetic resonance imaging was not performed due to the limitation of investigations in the state health sector. Her nerve conduction study showed evidence of ulnar nerve compression at the elbow supporting the diagnosis of ulnar nerve compression by the mass of calcium deposition.

Surgical exploration was performed, and significant ulnar nerve compression was noted due to the mass effect of the calcium deposition (Figure 2). Excision of the mass and ulnar nerve decompression with anterior transposition was performed as complete excision of the calcium deposition was not feasible and anterior transposition facilitated the final position of the nerve in a relatively disease-free tissue. Histology revealed a dark basophilic homogeneous material

Table 1. Reported cases of ulnar nerve entrapment syndromes due to calcinosis cutis.

	Author	Year	Age (years), gender	Sub type of CC	Site
1	Özalay et al. ⁵	2006	52, male	Metastatic (chronic kidney disease)	Cubital tunnel
2	Özalay et al. ⁵	2006	24, male	Metastatic (chronic kidney disease)	Cubital tunnel
3	Özalay et al. ⁵	2006	22, male	Metastatic (chronic kidney disease)	Cubital tunnel
4	Koratala et al. ⁸	2018	79, female	Dystrophic (scleroderma)	Cubital tunnel
5	Thurman et al. ⁶	1991	70, female	Dystrophic (scleroderma)	Guyon's canal
6	García et al. ⁷	2000	70, female	Metastatic (chronic kidney disease)	Guyon's canal

CC: calcinosis cutis.

with surrounding fibrosis suggestive of CC. Her post-operative recovery was uneventful with satisfactory symptom relief. There was no evidence of regrowth or recurrence of symptoms at 4 months of follow-up.

Discussion

Idiopathic CC is a diagnosis of exclusion where the patient does not exhibit elevated calcium levels (metastatic CC), history of tissue damage (dystrophic CC), and local extravasation of calcium (iatrogenic CC). In the reported patient, normal serum calcium levels and the absence of tissue trauma or features to suggest connective tissue disorder point to the diagnosis of idiopathic CC. The usual presentations of CC are painful subcutaneous nodules, extrusion of calcium deposits, and ulceration. Tissue dysfunction due to calcium deposition is rare, and nerve compression and airway dysfunction have been reported.¹

Several options in managing CC have been described in addition to treating the underlying autoimmune condition or biochemical abnormality. Topical or intralesional sodium thiosulfate, extracorporeal shockwave lithotripsy, and laser therapy have been utilized.² However, the utility of the above treatment options has not been described in treating nerve entrapment syndromes. Two studies including 11 and 39 patients, respectively, have described the surgical treatment of CC in the upper limb.^{3,4} All were painful nodules or ulcerations and managed successfully with good post-operative pain relief. None of the patients had symptoms of nerve entrapment.

Only six cases of ulnar nerve entrapment syndromes due to CC have been previously reported (Table 1⁵⁻⁸). Four patients had entrapment at the cubital tunnel and the other two at Guyon's canal. All had a background predisposition for calcium (either scleroderma or calcium dysregulation due to chronic kidney disease). All patients were managed with surgical excision of the calcium deposition with successful symptom relief. The type of surgical excision depends on the morphology of the calcified mass. In previously reported cases, four had well-circumscribed masses which were completely excised.⁵⁻⁷ Two patients had ill-defined

lesions, and one was treated with partial excision⁵ and the other one was managed conservatively due to lack of fitness for surgery.⁸ The reported patient is the first case to report ulnar nerve compression at the cubital tunnel due to idiopathic CC. Objective evidence of the ulnar nerve entrapment was determined by the nerve conduction study.

Conclusion

We report a rare case of idiopathic CC causing cubital tunnel syndrome. Although CC causing cubital tunnel syndrome has been reported, all patients had some form of calcium dysregulation. Therefore, the reported patient is the first case to report ulnar nerve compression at the cubital tunnel due to idiopathic CC. Excision of the mass and ulnar nerve decompression with anterior transposition was successful in our patient despite the incomplete excision of the calcium deposition.

Author contributions

O.B., U.J., and T.B. contributed to collection of information and writing of the manuscript. T.B. contributed to writing and final approval of the manuscript. All authors read and approved the final version of the manuscript.

Availability of data and material

All data generated or analyzed during this study are included in this published article.

Declaration of conflicting interests

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from the patient for anonymized information and accompanying images to be published in this article. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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