



**A soft tissue pseudotumor.
A case report of calcinosis cutis and literature review**

**pseudotumeur des parties molles.
À propos d'un cas de calcinose cutanée et revue de la littérature**

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ABSTRACT

Calcinosis cutis, an uncommon disorder, may be classified as idiopathic, metastatic or dystrophic. It has been often described in adults. Here we describe a case of a neonate who developed a dystrophic calcinosis cutis of her left ankle few days after receiving calcium gluconate by an intravenous administration for hypocalcaemia. The patient was addressed to our department with a suspected diagnosis of tumoral calcinosis, a rare benign soft tissue pseudotumor. Pathophysiological mechanisms, the course, prevention and differential diagnosis of this condition are discussed.

RÉSUMÉ

La calcinose cutanée, affection rare et souvent décrite chez l'adulte, peut être classée en idiopathique, métastatique ou dystrophique. Nous rapportons un cas ici le cas d'un nouveau-né qui a développé une calcification sous-cutanée de la face externe de la cheville gauche quelques jours après avoir reçu du gluconate de calcium en intraveineux pour une hypocalcémie néonatale. Le patient nous a été adressé pour une calcinose tumorale, une pseudotumeur rare des parties molles. A travers cette observation et une revue de la littérature, nous allons discuter la physiopathologie, l'évolution, les diagnostics différentiels et la prévention de cette affection.

KEYWORDS

calcinosis cutis, calcium gluconate, complication, pseudotumor, tumoral calcinosis

MOTS CLÉS

calcinose cutanée, gluconate de calcium, complication, pseudotumeur, calcinose tumorale

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INTRODUCTION

Calcinosis cutis, the cutaneous deposition of calcium salts in the dermis is an uncommon disorder. Occurring through a variety of pathogenic mechanisms, calcinosis cutis may be classified as metastatic, dystrophic or idiopathic and has been described rarely in the pediatric age group. We report calcinosis cutis in a term newborn who developed a convulsive disorder secondary to hypocalcaemia.

CASE REPORT

A 5-week-old female infant was referred to our department for a swelling and erythema of her left ankle. She had been treated for convulsive disorder in a neonatology department. Four days prior to the appearance of erythematous induration, the infant had received a 10% calcium gluconate intravenous infusion through the lateral saphenous vein at the ankle level of the same leg for the management of convulsions. The patient was then addressed to our department with a suspected diagnosis of tumoral calcinosis, a rare benign soft tissue pseudo-tumor.

The infant was reluctant to move her left lower limb. She was well developed and not febrile. Local examination revealed swelling and erythematous induration involving the lateral aspect of the left ankle (Fig 1). The swelling was warm and tender with central fluctuation. The regional lymph nodes were not enlarged.



Figure 1 Lateral erythematous swelling of the left ankle

Biology was normal [WBC = 7800/mm³; ESR = 13 (H1); CRP < 6mg/l].

Radiographs of the left ankle showed extensive extra-osseous and extra-articular calcification in the lateral aspect of the ankle region (Fig 2a & 2b) with a perivascular line calcification (Arrows - Fig 2b).

A diagnosis of iatrogenic calcinosis cutis was made. The child was treated symptomatically with antiinflammatory medication and the ankle was placed in a posterior splint. One week after, we noted a decreasing of local symptoms but the patient was lost to follow-up.

DISCUSSION

Deposition of calcium into tissues other than osteoid

2a Soft tissue calcification in the lateral extra-articular aspect of the ankle
2b Perivascular calcification (arrows)

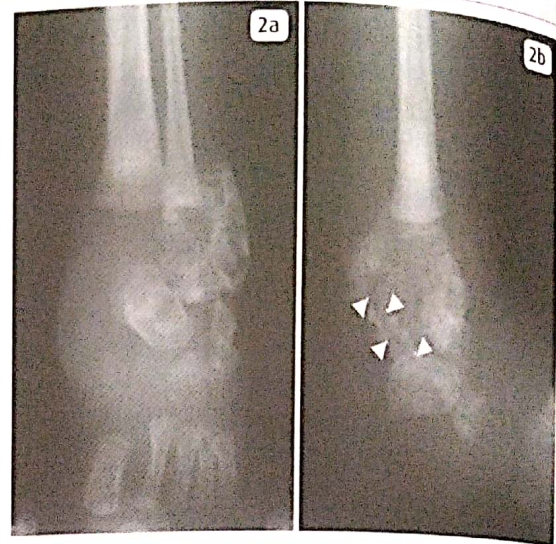


Figure 2 Left ankle X-rays

is known as heterotopic calcification [1]. Heterotopic calcification may be subclassified into metastatic calcification with a derangement in calcium metabolism, dystrophic calcification with normal serum calcium and phosphate levels and idiopathic calcification when the underlying pathophysiological mechanism is unknown.

Metastatic calcification can accompany systemic hypercalcaemia and may complicate primary or secondary hyperthyroidism, destructive lesions of bones and chronic renal failure [2]. Mostly, this presents in central areas.

Idiopathic calcification can take two forms: subepidermal calcified nodules and congenital calcinosis cutis. Subepidermal calcified nodules are usually solitary lesions which develop on the head or the extremities of young children. Without evidence, it has been suggested that the calcification occurs in a pre-existing nevus [3]. The congenital calcinosis cutis has a generalized distribution and the etiology is uncertain. In some cases, there is an association with Down's syndrome [4].

Dystrophic calcification generally occurs in areas of damaged or degenerate tissues which may be due to trauma, inflammation or neoplasia. Systemic diseases, such as dermatomyositis or systemic lupus erythematosus, can lead to such cutaneous deposit of calcium [5, 6].

The dystrophic calcification can follow extravasation of injected calcium solutions. The increase in the local concentration of calcium as well as tissue damage at the site of extravasation, contribute to the pathogenesis [7]. In fact, local tissue injury increases cell membrane permeability, allowing cytosolic influx of calcium that exceeds the capacity of mitochondria to sequester calcium and phosphate. This leads to the precipitation of calcium phosphate in the cytoplasm [8]. Mast cells might play a significant part because histamine and serotonin have been found to induce local calcification [9].

The final pathway of calcification is the formation of crystalline and insoluble calcium phosphate mineral, in the form of hydroxyapatite. Calcification occurs several days after the injury, appearing as firm nodules. Then appear oedema, erythema, induration and sometimes fluctuation or skin necrosis [10].

Skin necrosis, which is the most serious complication, is due to penetration of highly soluble calcium in the upper dermis where it is converted to insoluble calcium sulfate, carbonate, and phosphate, which in turn initiate a fibroblastic reaction and ultimately lead to coagulation necrosis [11].

Involvement of peripheral nerves in an extravasation injury is also reported [10, 12]. Tuncer described a case of calcinosis cutis surrounding the dorsal cutaneous branch of the ulnar nerve that justified surgical management [10].

Radiological changes become obvious one to three weeks later and may take any of three forms: amorphous masses localized to the site of extravasation with associated periosteal reaction [10], diffuse subcutaneous plaques [13] and vascular or perivascular line calcification [14]. In our case, we have noted the association of the two latter forms.

The diagnosis is frequently mistaken for cellulites, osteomyelitis, septic arthritis, abscess, myositis ossificans, and phlebitis [15]. Some patients were subjected to incision and drainage under anesthesia with a false diagnosis of abscess [16].

The diagnosis of iatrogenic calcinosis cutis rests on elucidation of the history of intravenous calcium solution along with radiological evidence of soft-tissue calcification, which is usually present when the local pathological signs are noticed [13]. When fluctuation is present, needle aspiration may be performed to differentiate the diagnosis from a pyogenic cause.

There is no specific treatment available for this condition. Fortunately, further course of calcinosis cutis is benign and treatment remains supportive therapy including elevation of the extremity, antiinflammatory medications and warm soaks. In most reported cases, progressive clearing of calcification starts occurring without any special treatment at about 8 weeks after the onset. At about 6 months, there is no evidence of tissue calcification [16, 17]. Treatment of calcification of the tissues by sodium sulphate or cellulose phosphate has been proposed [18? 19]. However, these methods are not suitable for use in infants [20]. In experimental situations, injection of intralesional triamcinolone acetate has been shown to reduce local signs [21].

Skin necrosis, if present, requires debridement and sometimes subsequent skin grafting [9]. There are several recommendations for reducing the risk of iatrogenic calcinosis cutis. Oral calcium supplementation should be used and preferred when possible. If intravenous administration is needed, it should not exceed 2ml/min

and gluconate is preferred to chloride. Adjunction of bicarbonates, phosphates or sulfates should be avoided. Cannulation sites should be rotated regularly and each cannula should be checked for a blood backflow before the infusion of calcium [22].

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