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CURRENT PROBLEM CASE

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Compression of the ulnar nerve in Guyon's canal by uremic tumoral calcinosis

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Abstract We describe the case of a 70-year-old woman with chronic renal failure on haemodialysis presenting with an ulnar nerve compression in Guyon's canal secondary to uremic tumoral calcinosis. Excision of calcium deposits and external neurolysis of the ulnar nerve were successfully performed. Simultaneously, the hyperphosphatemia and hypercalcemia were corrected. The pathogenesis of this condition is different from primary tumoral calcinosis. Clinical and radiological features and therapy are discussed. Uremic tumoral calcinosis is an unusual etiology of ulnar nerve compression in Guyon's canal not previously reported in dialysis patients.

Introduction

Compression of the ulnar nerve at Guyon's canal is an uncommon cause of cubital tunnel syndrome. The most important causes are rheumatoid disease or synovitis of the sheath of flexor carpi ulnaris. Other causes include muscle anomalies, lipoma, tumours, ganglion, ulnar artery thrombosis or aneurysm, repetitive trauma or fracture of the hamate bone [5]. We report an unusual case of ulnar nerve compression in Guyon's canal by uremic tumoral calcinosis.

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Case report

A 70-year-old woman with chronic renal failure due to bilateral renal lithiasis had been on haemodialysis since June 1993. The patient required medical attention because of the appearance of round, slowly growing tumours on the anterior surface of the right elbow, third finger of the right hand, anterocubital surface of the left wrist, palm of the left hand and second phalange of the second finger of the left hand. The patient also complained of paraesthesia in the fourth and fifth fingers of the left hand. Physical examination disclosed that the tumours were of firm consistency and painless when touched. Tinel's sign in the left wrist was positive.

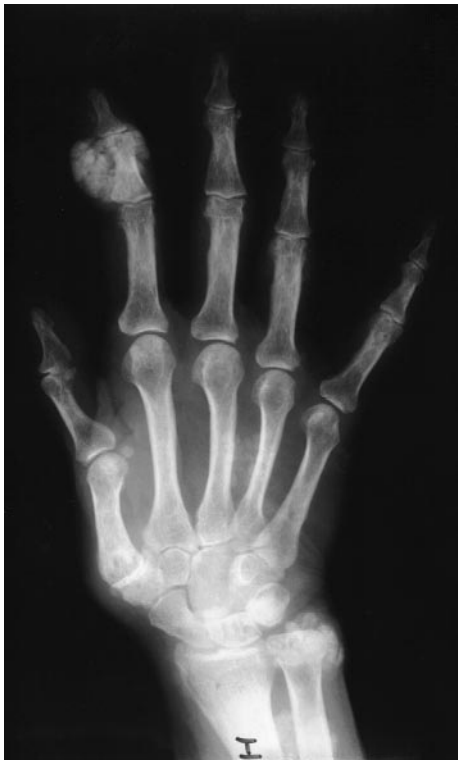

Fig. 1 Radiograph of the left hand showing lobulated, calcified masses on the anterocubital surface of the wrist, between the metacarpals and second phalange of the second finger

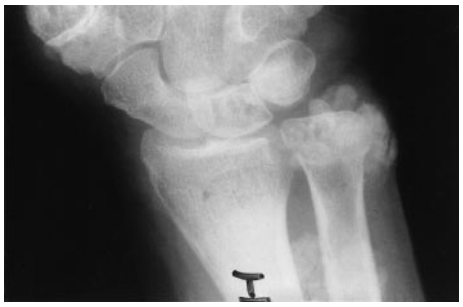

Fig. 2 Detail of the left carpus displaying a nodular calcified mass at Guyon's canal

Plain radiographs revealed a lobulated, homogeneous, densely calcified periarticular mass, not involving the joint spaces in the sites previously described. The involvement of the anterocubital surface of the left wrist was located at the level of Guyon's canal (Figs. 1 and 2). Electrophysiological study confirmed the compression of the cubital nerve at the level of the wrist. Laboratory data revealed calcium 10.8 mg/dl (2.7 mmol/l), phosphorous 6.8 mg/dl (2.2 mmol/l), alkaline phosphatase 127 U/l and parathyroid hormone 98 ng/l (normal value < 60 ng/l). The retrospective evaluation of calcium-phosphorous metabolism showed a very high calcium-phosphorous product (Ca × P) persistently above 70 (measurements in conventional units), due to a hypercalcemia and an uncontrolled hyperphosphoremia caused by the continuous administration of calcium carbonate and oral calcitriol and a dietary excess of phosphorus. Severe hyperparathyroidism was excluded.

Calcifications in the wrist, palmar surface and second finger of the left hand were removed by surgery. An external neurolysis was performed because the tumoral calcinosis was compressing the deep motor branch of the cubital nerve. The lesions, ranging between 2 and 3 cm in diameter, were encapsulated and contained a creamy toothpaste-like material. Microscopy showed numerous calcified masses surrounded by a granulation tissue with histiocytes and some multinucleated cells. Tumoral calcinosis was diagnosed. Simultaneously, the duration of dialysis was increased, the hyperphosphoremia was corrected, and calcitriol was suspended, thereby obtaining a reduction of the calcium-phosphorous product. Paresthesia disappeared in the early period after surgery, and an electrophysiological study conducted 3 months later was normal. No recurrence of the tumoral calcinosis was documented after a 14 month follow-up.

Discussion

Tumoral calcinosis is an unusual and benign condition characterized by large, calcified, periarticular soft-tissue masses composed of calcium salts, and usually located around large joints. There is a primary form (idiopathic or hereditary form), but it may also be found in a wide variety of conditions: primary or secondary hyperparathyroidism, vitamin D intoxication, scleroderma or uremic tumoral calcinosis [6, 7].

Tumoral calcinosis is a condition observed with increasing frequency in patients with chronic renal failure on dialysis. The pathogenesis is not well understood. Precipitation of calcium phosphate salts occurs when the solubility of the calcium-phosphorus product reaches a critical value (usually Ca × P > 70). Classically, the most important cause was severe hyperparathyroidism. However, at present it is more frequent in the absence of secondary hyperparathyroidism, due to the increase in the calcium-phosphorus product caused by iatrogenic hypercalcemia (administration of calcitriol or calcium carbonate) or by persistent hyperphosphoremia [2].

The lesion consists of encapsulated and multilobulated masses, of variable size, containing a creamy toothpaste-like material. The clinical manifestations are periarticular round tumours with a firm consistency, progressively growing and usually located around the large joints [3]. Radiographs revealed lobulated, homogeneous, densely calcified periarticular masses, usually around the large joints, and with normal joint spaces. Computed tomography can disclose the presence of fluid-calcium levels (sedimentation sign), and magnetic resonance images display low signal density on T1- and T2-weighted sequences. Radiologic characteristics allow tumoral calcinosis to be distinguished from other diseases which produce soft-tissue calcification. Nevertheless, a biopsy can be warranted when the diagnosis is doubtful, and a musculoskeletal tumour must be excluded [6, 7].

The treatment of uremic tumoral calcinosis consists of the correction of the factors responsible for the calcium-phosphorus product elevation. Surgery is advised when the masses cause symptoms. Renal transplant and an increase of dialysis have produced a reduction of the masses by inducing a negative calcium balance [2].

In patients undergoing long-term dialysis, the development of carpal tunnel syndrome and, less frequently, cubital nerve compression due to dialysis- related amyloidosis (ß2-microglobulin deposit) is well-known [4, 9]. However, nerve compression due to tumoral calcinosis is uncommon in dialysis patients. Two cases of tumoral calcinosis causing compression of the ulnar nerve in Guyon's canal have been reported in patients with systemic scleroderma, but their renal function was normal [1, 8]. On the other hand, a carpal tunnel syndrome caused by tumoral calcinosis was described in another patient without renal disease [10].

In conclusion, uremic tumoral calcinosis is an infrequent etiology of ulnar nerve compression in Guyon's canal so far not reported in dialysis patients.

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