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Reconstructive

Tumoral Calcinosis: A Giant Tumoral Mass in the Shoulder Joint of a Dialysis Patient

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Summary: Tumoral calcinosis is a rare condition in which a calcified mass grows around a large joint, and can occur in patients undergoing renal dialysis. Here, we report the case of a 64-year-old man with a long history of dialysis who presented with a giant, painless mass in his right shoulder joint. A near-complete surgical resection is performed without muscle function loss and with no sign of recurrence after 1 year. *(Plast Reconstr Surg Glob Open 2018;6:e1730; doi: 10.1097/GOX.0000000000001730; Published online 9 April 2018.)*

CASE REPORT

A 64-year-old man with a 10-year history of a slow-growing mass without pain in his right shoulder joint that decreased the range of motion and induced numbness of his right hand with nighttime exacerbation was referred to our hospital. Puncture aspirations of the mass had been performed several times by his previous doctors, with the mass slowly relapsing each time. The patient also had a 20-year history of hemodialysis for hypertensive nephrosclerosis. Five years before, he had undergone parathyroidectomy and autoimplantation of his parathyroid tissue to his right forearm for secondary hyperparathyroidism. There was no history of trauma to these areas or a family history of pathological calcifying disorders (e.g., hyperphosphatemic familial tumoral calcinosis, gout, or pseudogout). A physical examination revealed firm, nontender, irregular-shaped nodules >20 cm in diameter. Laboratory assessments revealed a slightly increased serum phosphate level but normal serum calcium. Large, irregular, radiodense calcifications adjacent to the shoulder joint were observed by radiology, and three-dimensional computed tomography (CT) of the chest revealed a giant tumoral mass in the shoulder joint measuring 25×20 cm (Fig. 1) [**see figure**, **Supplemental Digital Content 1**, which displays preoperative chest x-ray showing an amorphous and multilobular calcification (blue arrow), ***http://links.lww.com/PRSGO/A716***]. Therefore, the preliminary diagnosis was tumoral calcinosis.

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The surgical findings showed that the mass was so big, that it even infiltrated to his axillary region and was localized under the latissimus dorsi and the serratus anterior muscles, which were subsequently dissected (Fig. 2) [**see figure**, **Supplemental Digital Content 2**, which displays preoperative CT scan showing a calcification of the axillary region under the serratus anterior muscle and latissimus dorsi muscle (blue arrow) ***http://links.lww.com/PRSGO/A717***]. To remove the mass from the posterior margin of the serratus anterior muscle, better access was achieved by splitting the muscle at the anterior part in the direction of the fiber while preserving its function. It was hard to access to the remnant of the mass in the axillary region by flexing the right upper arm with left lateral recumbent position. So, we widened the surgical field by changing the position of the right upper arm to extend to zero degree. We tried to excise the mass adherent to the ribs using rongeur forceps. However, portions of the mass that infiltrated the intercostal spaces were left to avoid causing pneumothorax when attempting to excise them by force, which can be seen on postoperative CT (Fig. 3).

Macroscopically, the excised mass was a 23×19×6 cm polycystic, calcified structure (**see figure**, **Supplemental Digital Content 3**, which displays macroscopic view of the excised mass, ***http://links.lww.com/PRSGO/A718***). When the mass was sectioned, a milky and sticky fluid was released. Hematoxylin and eosin staining showed extensive amorphous calcium deposits within a fibrous stroma calcification (Fig. 4), between which were giant multinucleated foreign-body cells and inflammatory infiltrates. No malignant cells were found, thus confirming the diagnosis of tumoral calcinosis.

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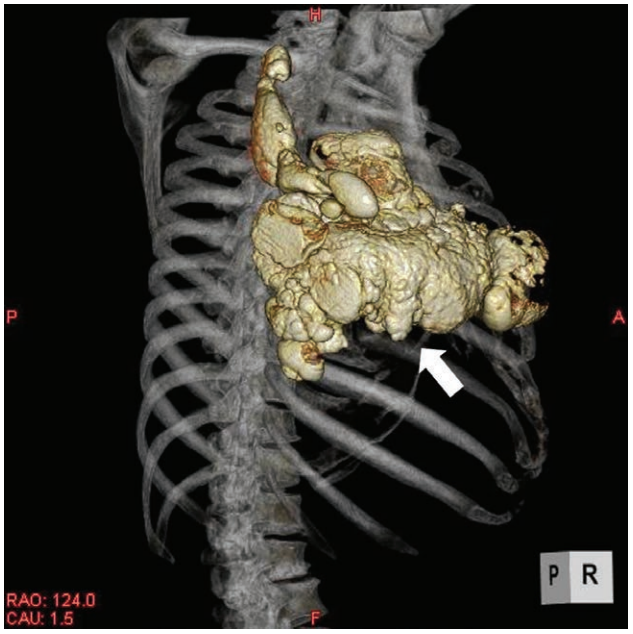

Fig. 1. Preoperative 3D CT scan demonstrating a large mass around the shoulder joint (white arrow).

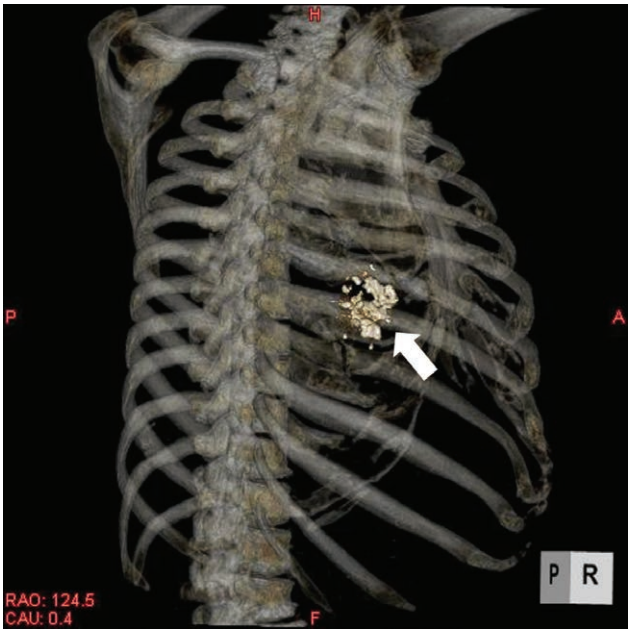

Fig. 3. Postoperative 3D CT scan showing the remnant of the mass in the intercostal area (white arrow).

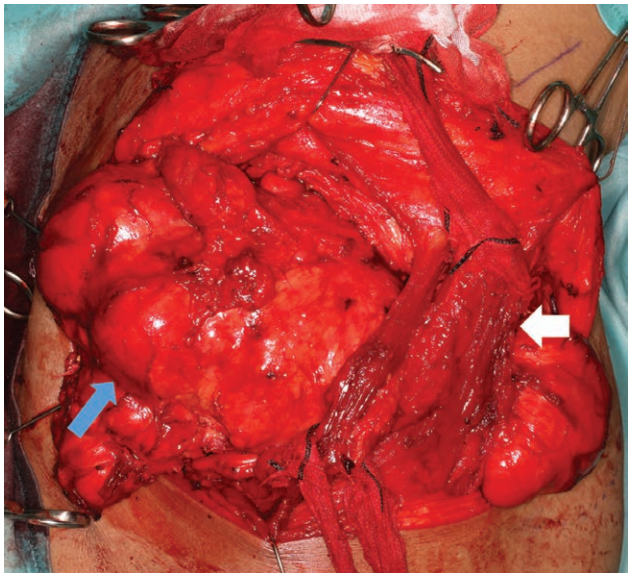

Fig. 2. Intraoperative image showing the large mass (blue arrow) under the serratus anterior muscle (white arrow).

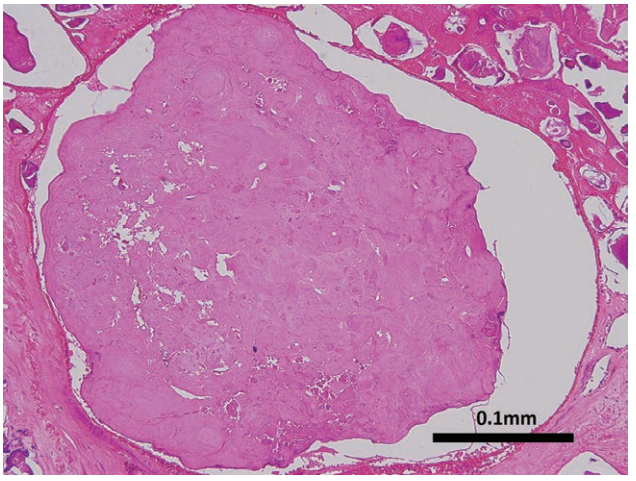

Fig. 4. Basophilic materials (calcification) were found in the resected tissues. Hematoxylin and eosin, ×100 magnification.

After 1 year, the patient had an improved range of motion of the shoulder joint and numbness of the hand, with no sign of hyperphosphatemia or new calcifications around the shoulder on a follow-up assessment.

DISCUSSION

Tumoral calcinosis is a rare disease characterized by subcutaneous soft tissue deposits of calcium phosphate near large joints, particularly the trochanteric and gluteal areas of the hip, the lateral portion of the shoulder, and the posterior elbow.[1] The prevalence of tumoral calcinosis in hemodialysis patients has been reported to range from 0.5% to 1.9%.[2,3] First described in the late 1890s by Giard[4] and Duret,[5] the term "tumoral calcinosis" was introduced in 1943 by Inclan et al.[6] Tumoral calcinosis is considered primary or secondary according to the presence or absence, respectively, of the underlying disease. For example, soft tissue calcification is considered secondary tumoral calcinosis in patients with concurrent chronic renal failure, pseudoxanthoma elasticum, malignancy, sarcoidosis, primary hyperparathyroidism, scleroderma, hypervitaminosis D, milk-alkali syndrome, or massive osteolysis. In the case presented here, the patient had a long history of renal dialysis with no family history of tumoral calcinosis, leading us to suspect secondary tumoral calcinosis.[7]

Although the pathogenesis of tumoral calcinosis is not known, repeated tissue injuries and inflammation seem to

play a role in initiating the calcifying process. For example, hemorrhaging or fat necrosis can lead to fibrosis with chronic inflammation, followed by massive calcification.[1,8] Tumoral calcinosis is primarily treated by surgically removing the lesions as early as possible, when they are small and amenable to total resection, followed by phosphate deprivation to prevent relapse. If incompletely excised, the residual mass can recur with a more rapid growth rate.[1,7] In cases complicated by secondary hyperparathyroidism, subtotal or total parathyroidectomy should be considered.[10] In this case, however, the serum phosphate level is not necessarily too high to be deprived of by medications, due to the patient's history of parathyroidectomy. Therefore, tumoral calcinosis patients, including this case, should be checked regularly to keep their serum phosphate levels low, and radiologically assessed for recurrence of tumoral calcinosis as a follow-up.

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