## Case Report

DOI: 10.5455/2349-2902.isj20150219

# Large tumoral calcinosis and pathological femur fracture in a hemodialysis patient with secondary hyperparathyroidism

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Received: 12 November 2014 Accepted: 9 December 2014

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#### **ABSTRACT**

Tumoral Calcinosis (TC) is a rare complication of uremic patients on dialysis. Bone erosion or fracture rarely happens in TC patients. TC is easy to be misdiagnosed as a malignant bone tumor by inexperienced orthopedists. We report the clinical, radiological and magnetic resonance imaging findings in a rare case of large TC which caused femur erosion both and fracture in a 49-year-old woman with uremia and secondary hyperparathyroidism (HPT) on hemodialysis and describe the surgical treatment and pathology results.

Keywords: Tumoral calcinosis, Pathological fracture, Hemodialysis, Secondary hyperparathyroidism

## INTRODUCTION

TC is a rare pathological entity and is characterized by calcium-phosphate crystals within the subcutaneous soft tissues around large joints. Uremic TC (UTC) is the term for TC occurring in uremic patients. 1,2 UTC rarely induces bone erosion or fracture.3 To the best of our knowledge, only 5 cases with bone erosion or fracture have been reported in the previous published literature.<sup>3-7</sup> Here, we present a very rare case of UTC with femur erosion both and fracture.

### **CASE REPORT**

A 49-year-old woman was admitted to our hospital with a swelling over the left proximal thigh. The swelling has existed for 2 years. Her left hip joint has not moved freely for 15 days before her hospitalization without obvious precipitating factors. She has received thrice weekly maintenance hemodialysis for chronic renal failure for 12 years. She denied the history of familial TC, infection, and major traumatic injury. Additionally, no masses were found around other joints. Physical examination revealed a large and hard mass around the left proximal thigh. The patient complained a painful sensation while palpating the mass. The overlying skin and local temperature was normal. The left hip joint could not move freely.

laboratory evaluation revealed: total alkaline phosphatase 726 U/L (reference range 35-100 U/L), blood urea nitrogen 15.45 mmol/L (reference range 2.95-7.70 mmol/L), creatinine 526 µmol/L (reference range 37-110 µmol/L), calcium 2.42 mmol/L (reference range 2.1-2.7 mmol/L), phosphorus 1.74 mmol/L (reference range 0.81-1.45 mmol/L), and parathyroid hormone (PTH) 169.2 pmol/L (reference range 1.60-6.90 pmol/L).

Anteroposterior (AP) radiograph showed a large mass with homogeneous, calcified density around the left proximal femur (Figure 1A). Magnetic Resonance Imaging (MRI) showed the lesion with hypo-intense and

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the erosion of underling cortex and marrow cavity of the left proximal femur on T2-weighted image (Figure 2).

serial examinations and consultations departments of thyroid surgery, nephrology, and anesthesiology, a definite diagnosis of UTC, left femur pathological fracture, uremia and secondary HPT was made. The consultation recommended parathroidectomy first, the second for orthopedic operation. The patient did not receive parathyroidectomy first and wanted to recover the motion of left hip joint as soon as possible, although she was told for many times that TC has the possibility to shrink or even disappear spontaneously parathyroidectomy. Therefore, orthopedic operation was performed first when the patient signed a consent form of operation. The lesion of TC and the left proximal eroded femur were completely resected; besides, the partial hip arthroplasty was performed with a proximal femoral endoprosthesis.

At surgery, a large, ovoid, hard, focally nodular mass was noted, about  $15.5 \times 15 \times 11$  cm in size, around the left proximal femur. The external surface was variably smooth and intact. Upon sectioning, the specimen was a cystic lesion containing massive yellow-gray chalky materials; besides, femur erosion and fracture was also found (Figure 3).

Pathology results confirmed the diagnosis of TC. Hematoxylin-Eosin (H&E) staining indicated bluish nodules which suggested the deposits of calcium salts; meanwhile, fibrous connective tissues were also found between these nodules which were surrounded by the osteoclast-like giant cells, histiocytes, but no malignant cells, with a clear edge and a foreign body-type granulomatous reaction (Figure 4).

The patient received the total parathyroidectomy with ante-brachial subcutaneous autotransplantation at one month after orthopedic operation. According to the physician's advice, low-phosphate dietary, hemodialysis, and other nonsurgical treatments should be continued after surgery. Post-operative recovery was uneventful (Figure 1B). No clinical and radiological signs of recurrence were seen at 10-month follow-up.



Figure 1: Plain radiograph (A): AP radiograph shows a multilobulated calcified mass around the left proximal femur. (B): AP radiograph shows the postoperative appearance of the left thigh.



Figure 2: The sagittal view of T2-weighted MRI demonstrates the mass with hypo-intense and the erosion of femur cortex and marrow cavity with hyper-intense.



Figure 3: Sectioning appearance of the specimen dissected from the femur lesion shows massive yellow-gray chalky materials and femur erosion and fracture.

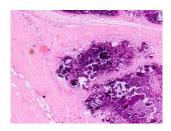


Figure 4: Microscopic appearance of the specimen dissected from the femur lesion shows bluish nodules surrounding fibrous connective tissues (H&E, x100).

## **DISCUSSION**

Hemorrhage The prevalence of UTC ranges from 0.5% to 3% in dialysis patients. <sup>1,8,9</sup> The most common sites are the large joints such as the hip, the elbow and the shoulder. <sup>1</sup> UTC is often a periarticular and large lesion, about 2-10 cm in diameter. <sup>8</sup> In our patient, the most diameter of the mass was up to 15.5 cm.

TC is uncommon benign entity and rarely induces bone erosion.<sup>3</sup> But bone erosion or even fracture may happen in uremic patients on long-term dialysis. By performing a PubMed literature search, we were able to identify 5 previous reported cases of UTC with bone erosion or fracture (Table 1). There was no gender difference among the 6 cases, including our case. The average age was 58.3years (range 43-84 years). The location was mostly

limb bones (4 cases), the rest was spine. And the average time of dialysis was 9.2 years (range 2-16 years).<sup>3-7</sup> Age,

lesion location and dialysis time of our patient wholly conformed to the ranges of previous reports.

Table 1: Total six cases of uremic tumoral calcinosis with bone erosion or fracture.

References	Patient data				Dialysis	Bone erosion or fracture
Author	Year	No.	Age	Gender	time	bone erosion or fracture
Meltzer et al.	1992	1	73 y	F	7 y	The left iliac crest
Huang et al.	2006	2	54 y	M	6 y	The proximal humerus
Ureten et al.	2008	3	43 y	F	16 y	The right femur neck
Demetriades et al.	2011	4	47 y	F	2 y	Thoracic vertebral bodies (T8-9)
Faur et al.	2013	5	84 y	M	12 y	Cervical vertebral l bodies (C1-2)

M: Male; F: Female; y: years

The etiology of bone erosion and fracture caused by UTC is not fully understood. At present, there are two predisposing factors. Firstly, secondary HPT may be the most important factor. In secondary HPT, PTH levels may be significantly increased and are frequently higher than the levels in primary HPT. PTH can stimulate bone resorp-tion which leads to osteoporosis. Secondly, the large size of the mass and its periarticular location induces microtrauma of the adjacent bone. The repeated microtrauma causes pressure erosion. Spontaneous fracture may eventually occur in the osteoporotic and eroded bone on certain situation.

It can be easily misdiagnosed by inexperienced orthopedists, because many periarticular masses may mimic the presenting pictures of TC, such as osteosarcoma, chondrosarcoma, myositis ossificans or heterotopic bone. 10 TC is the benign entity without malignant cells under light microscope. Biopsy is necessary when the lesion is difficult to diagnosis. Pathology examination is the gold standard for the diagnosis of TC. Medical and family history, physical examinations, imaging tests, laboratory examinations, and pathological examinations should be assessed carefully and comprehensively before making the definite diagnosis.

The mainstay treatments of UTC include the two following aspects. On the one hand, nonsurgical treatments include dietary phosphorus restriction, noncalcium phosphate binders, calcimimetics, optimal control of secondary HPT, and intensive hemodialysis with low-calcium dialysate. 1,8,9 But the rate of success is different due to different factors. On the other hand, operations surgical include lesion resection, parathyroidectomy and renal transplantation. Surgical operations are recommended for persistent or refractory UTC. It is important to note that surgical operations accompany a variable rate of recurrence. 1,8,9 In our patient, there was no relapse at the latest follow-up. It is necessary to comprehensive consider both of nonsurgical

treatments and surgical operations, according to the different conditions of patients.

Funding: No funding sources Conflict of interest: None declared

Ethical approval: The study was approved by the hospital

ethics committee

#### REFERENCES

- 1. Shpilberg KA, Blowe SE, Som PM. Mass-like and extensive secondary tumoral calcinosis in the neck and body of a patient on peritoneal dialysis. Clin Imaging. 2013;37(5):972-5.
- 2. Olsen KM, Chew FS. Tumoral calcinosis: pearls, polemics, and alternative possibilities. Radiographics. 2006;26(3):871-85.
- 3. Huang YT, Chen CY, Yang CM, Yao MS, Chan WP. Tumoral calcinosis-like metastatic calcification in a patient on renal dialysis. Clin Imaging. 2006;30(1):66-8.
- 4. Meltzer CC, Fishman EK, Scott WW Jr. Tumoral calcinosis causing bone erosion in a renal dialysis patient. Clin Imaging. 1992;16(1):49-51.
- Ureten K, Oztürk MA, Ozbek M, Unverdi S. Spontaneous and simultaneous rupture of both Achilles tendons and pathological fracture of the femur neck in a patient receiving long-term hemodialysis. Int Urol Nephrol. 2008;40(4):1103-6.
- 6. Faur D, Pérez-Bovet J, Ferrer SM, Prats MV. Pathologic cervical fracture in a patient with secondary hyperparathyroidism. Kidney Int. 2013;83(5):974.
- Demetriades A, Wong F, Ellamushi H, Afshar F, Yeh J. Balloon kyphoplasty treatment for a spontaneous vertebral fracture in renal osteodystrophy. BMJ Case Rep. 2011;2011: pii: bcr0220113890.
- 8. Younes M, Belghali S, Zrour-Hassen S, Béjia I, Touzi M, Bergaoui N. Complete reversal of tumoral calcinosis after subtotal parathyroidectomy in a

- hemodialysis patient. Joint Bone Spine. 2008;75(5):606-9.
- 9. Chu HY, Chu P, Lin YF, Chou HK, Lin SH. Uremic tumoral calcinosis in patients on peritoneal dialysis: clinical, radiologic, and laboratory features. Perit Dial Int. 2011;31(4):430-9.
- 10. Mohamed S, Jong-Hun J, Weon-Yoo K. Tumoral calcinosis of the foot with unusual presentation in

an 11-year-old boy: a case report and review of literature. J Postgrad Med. 2007;53(4):247-9.

DOI: 10.5455/2349-2902.isj20150219 **Cite this article as:** Liu X, Hong S, Chen G, Tu C. Large tumoral calcinosis and pathological femur fracture in a hemodialysis patient with secondary hyperparathyroidism. Int Surg J 2015;2:91-4.