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

Xiwei Liu¹, Song Hong¹, Ganjun Chen², Chongqi Tu¹*

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 published previous literature.³⁻⁷ 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 femur. The patient complained a painful sensation while palpating the mass. The overlying skin and local temperature were normal. The left hip joint could not move freely.

ABSTRACT

Tumoral Calcinosi(s) (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

1Department of Orthopedics, West China Hospital, Sichuan University, Chengdu, China
2Department of Pathology, West China Hospital, Sichuan University, Chengdu, China

Received: 12 November 2014
Accepted: 9 December 2014

*Correspondence:
Dr. Chongqi Tu,
E-mail: chongqitu@163.com

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DOI: 10.5455/2349-2902.isj20150219
the erosion of underling cortex and marrow cavity of the left proximal femur on T2-weighted image (Figure 2).

After serial examinations and consultations of 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 parathyroidectomy 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 after 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 × 15 × 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.

**DISCUSSION**

Hemorrhage The prevalence of UTC ranges from 0.5% to 3% in dialysis patients. The most common sites are the large joints such as the hip, the elbow and the shoulder. UTC is often a periarticular and large lesion, about 2-10 cm in diameter. 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. 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 58.3 years (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). \(^3\) 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.

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

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 resorption which leads to osteoporosis. \(^2\) Secondly, the large size of the mass and its periarticular location induces microtrauma of the adjacent bone. The repeated microtrauma causes pressure erosion. \(^4\) 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, non-calcium 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, surgical operations 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

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DOI: 10.5455/2349-2902.isj20150219