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Case Report

Conservative Treatment in Impending Fracture of Solitary Brown Tumor of Tibia: A Case Report and Review of Literature

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Abstract

Background: Brown tumors, also known as osteitis fibrosa cystica, are focal benign bone lesions caused by an increased osteoclastic activity because of high levels of parathyroid hormones. The increase of parathyroid hormones can be primary hyperparathyroidism (HPT) and secondary HPT resulting from renal failure and vitamin D deficiency. ***Case presentation:*** A 26-year-old Thai female visited the clinic because of right leg pain and swelling for three weeks. Plain radiography revealed a solitary osteolytic lesion with an impending fracture at the anterior cortex of the right proximal tibia. The patient underwent a core needle biopsy. The initial histopathology report depicted a giant cell lesion. In addition, the patient had elevated serum parathyroid hormones. A parathyroid adenoma was discovered on a further investigation with contrast-enhanced computed tomography of the neck. The immunochemistry and molecular study report were consistent with a brown tumor. The patient underwent parathyroidectomy for her adenoma and was on weight-bearing restrictions for her leg. After three months of follow-up, she could bear weight on her right leg without pain, and radiographic evaluation showed restoration of the tibial cortex. ***Conclusion:*** Conservative management has a role in managing brown tumors of the lower extremities with impending pathologic fracture. Rapid diagnosis and removing the primary cause of parathyroid adenoma as soon as possible is a crucial part of the treatment.

Keywords: ***Brown tumor, Hyperparathyroidism, Solitary Brown tumor, Bone metabolic disorder***

Background

Brown tumor, also known as osteitis fibrosa cystica, is one of the consequences of excessive parathyroid hormone levels. Hyperparathyroidism can occur because of intrinsic causes related to the parathyroid gland, commonly parathyroid hyperplasia or parathyroid adenoma [1]. Secondary causes include vitamin D deficiency and chronic kidney disease [2]. Parathyroid adenoma causes 80%-90% of primary hyperparathyroidism cases. The literature's reported incidence of brown tumors ranges from 1.5% to 4.5% [3].

Case Presentation

A 26-year-old female visited the hospital due to swelling and pain in her right leg. The patient did not have any family history of metabolic diseases. The patient presented to the outpatient department (OPD) with ongoing swelling and pain in her right leg for three weeks previously. The pain was aggravated while weight-bearing on the right leg. Tenderness and swelling at the proximal tibia of the right leg were noted. Plain radiography revealed multiple geographic intramedullary cystic lesions of the right proximal tibia. During the three weeks before the visit to our clinic, her pain was well-controlled with medication. She was ambulatory but with slight discomfort in her right leg. Physical examination showed a tender, poorly defined mass of 10 × 5 cm at the anterior aspect of the right proximal leg.

Right tibial plain radiography (**Fig. 1**) showed osteolytic lesions at the proximal shaft of the tibia and impending fracture of the anterior cortex of the tibia.

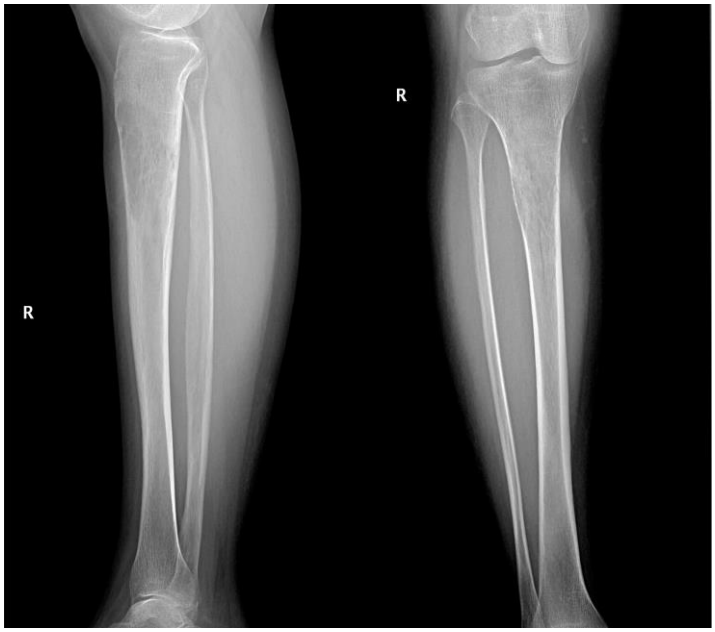

Figure 1: Pre-operative radiograph shows osteolytic lesion at a proximal shaft of the tibia with extraosseous extension, associated soft tissue component, and impending fracture of the anterior cortex of the tibia.

Magnetic resonance imaging showed a 3.1 × 3.8 × 8.6 cm marrow infiltrative mass involving proximal tibia, with cortical break-through and soft tissue extension at the anterior tibial cortex.

Serum blood chemistry showed an elevated parathyroid hormone level (407.90 pg/ml, normal range: 15-56), a total calcium level slightly above the normal upper limit (11.4 mg/dl, normal value: 8.6-10), and a depressed phosphorus level (2 mg/dl, normal value: 2.7-4.5). Serum creatinine was 0.64 mg/dl (normal value: 0.51-0.95). Contrast-enhanced computed tomography scans of the neck suggest parathyroid hyperplasia/parathyroid adenoma. The histopathology obtained by core needle biopsy at the right proximal tibia showed a giant bone-cell lesion. Immunohistochemistry was positive for SATB2 and osteocalcin and negative for S-100. The molecular study was negative for H3F3A mutation; thus, the diagnosis of brown tumor was preferred over giant cell tumor of the bone and giant cell-rich osteosarcoma.

The weight-bearing restriction was initiated. The patient was also given calcium and vitamin D supplements. An experienced surgeon was consulted to perform parathyroidectomy to remove the parathyroid adenoma's primary cause. We immediately noticed dramatic improvements when the patient came for her scheduled follow-up two weeks after surgery. The pain she felt in her right leg was substantially reduced, and she did not feel as distracted by the pain when performing her usual daily activities. At a later follow-up of three months, the pain gradually resolved, and she could bear weight on her right leg without any pain or discomfort. Plain radiography (Fig. 2) showed a gradual decrease in the osteolytic lesion.

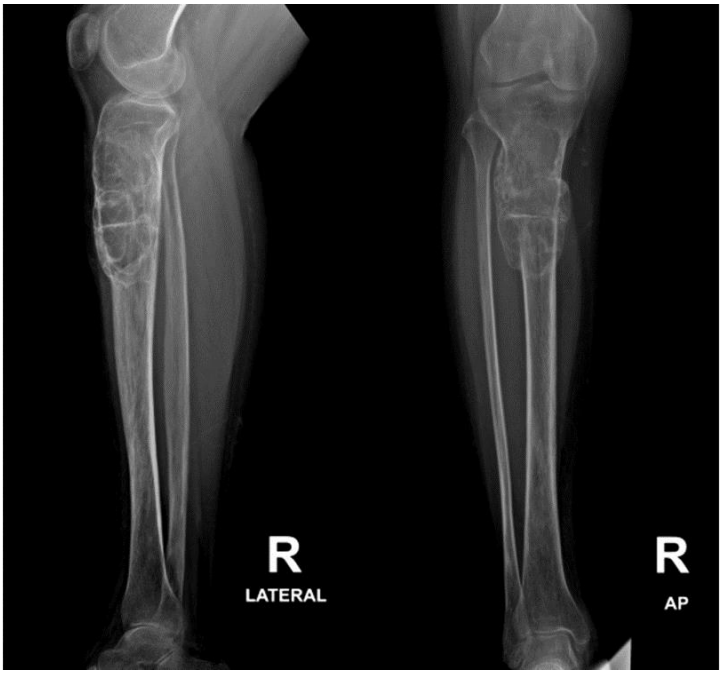

Figure 2 Plain film of post-treatment shows thickening anterior cortex of the tibia.

Serum parathyroid hormone level returned to the normal range (45.62 pg/ml, normal range: 15–65) from 2 weeks after surgery and remained within normal values.

Discussion and conclusion

This case reports a solitary brown tumor of the tibia, which poses a difficult diagnostic challenge for physicians. Table 1 shows the clinical, radiological, and histological features of giant cell lesions.

Table 1: Clinical, radiological, and histological features of giant cell lesion.

Disease	Type	Clinical	Radiographic characteristics	Histological features/immunohistochemistry/molecular study
Brown tumor	Non-neoplastic	Painful mass	Solitary or multiple expansile lytic lesion occasionally with intralesional trabeculae	Lobular architecture produced by fibrous septa that may contain trabeculae of reactive woven bone. The lobules are composed of an admixture of plump fibroblasts, extravasated red blood cells, hemosiderin-laden macrophages, and scattered osteoclast-type giant cells, which frequently cluster around areas of hemorrhage.
Giant cell tumor	Benign neoplasm	Frequently produces pain	Eccentric, lytic mass that frequently extends from the subchondral bone plate into the metaphysis. Larger tumors may involve the adjacent diaphysis and/or the neighboring soft tissues.	Large number of multinucleated osteoclast-like giant cells scattered throughout the tumor. The cell contains oval, vesicular nuclei with central nucleoli. The round or oval mononuclear stromal cells are the diagnostic and neoplastic component of the tumor. Immunohistochemically, the cells express vimentin and alpha-1-antitrypsin and do not stain with antibodies to S-100.
Giant cell-rich osteosarcoma	Malignant neoplasm	Painless swelling of extremities	Metaphyseal mixed lytic and blastic destructive mass with poorly defined margins	Histologically contains more than 50% of osteoclast-like cells mixed with malignant bone-forming cells. The malignant cells demonstrate significant cytological atypia (pleomorphism, hyperchromatic, high mitotic rate).

Focal osteolytic lesions of the bone may mimic primary bone malignancies, thus causing difficulties in diagnosis [4]. We found seven reported cases of brown tumor at the tibia in Table 2 [3,5-11].

Table 2: Reported cases of brown tumor at the tibia in literature.

Author	Age (years), sex	Clinical	Radiographic evaluation	Treatment	Outcome	Cause
Yalçin et al. (2017) [6]	12, M	Pathological fracture	Cystic lesions in the femoral supracondyle, left acetabular roof, and right proximal and distal tibia	Curettage and grafting with spongy allograft were carried out at the tibia and observation for other areas	Hip, ankle pain-free at 5 months. Tibia, able to start weight-bearing. No growth of the tumor was observed.	Secondary hyperparathyroidism because of renal osteodystrophy (On hemodialysis)
Al-Sharafi et al. (2015) [3]	50, F	Three months of painful swellings in the right hand and leg, difficulty walking	Osteolytic lesion in the right second metacarpal bone and a cortical osteolytic expansile lesion in the middle aspect of the right tibial diaphysis	Vitamin D3 drops 45,000 IU weekly and calcium 500 mg daily	Decreased pain, walking without cane at 3 months. ALP, calcium remained slightly elevated at 16 months.	Secondary hyperparathyroidism because of vitamin D deficiency
Vaishya et al. (2017) [5]	25, F	One month of right leg pain associated with difficulty walking	Geographical lytic lesion in the diaphysis of the distal one-third of the tibia with complete erosion of the posteromedial cortex	Vitamin D supplementation, and the leg was immobilized in a below-knee cast for 2 months	Tumor regressed. Clinical improvement. Lab results normal after 3 months.	Secondary hyperparathyroidism (Severe vitamin D deficiency due to breastfeeding)
Toriu et al. (2019) [7]	48, M	Pain in lower right leg	Spontaneous fracture of the right distal tibia. MRI show bone-tumor-like mass lesion associated with a fracture line	Open resection and internal fixation of the distal right tibia	Free from symptoms 2 years post-operatively	HPT resistant to medical treatment S/P PTX 3 years ago
Rueda et al. (2021) [8]	N/A	N/A	N/A	N/A	N/A	Primary hyperparathyroidism
Present et al. (1988) [9]	49, F	N/A	N/A	N/A	N/A	Renal osteodystrophy
Hori (1970) [10]	N/A	N/A	N/A	N/A	N/A	N/A
Sprenger-Mähr et	26, F	Bone pain at left hip	MRI show cystic lesion compatible	Right acetabulum cavities were filled	Second enucleations of brown tumor in the right	Secondary parathyroidism due

al. (2019) [11]			with a brown tumor at acetabulum, tibia, ribs, skull, thoracic spine, and thumb	with autologous and homologous bone graft. Osteosynthesis of the tibia. Etelcalcetide started 3 months after delivery.	acetabulum and in the right tibia became necessary 8 and 12 months after starting with etelcalcetide, because the bone grafts had been absorbed. New bone developed in the thoracic spine 2 years after starting etelcalcetide. After interventions the patient was fully mobile and without pain.	to pregnancy, ESRD, and lupus nephritis

Of the seven cases, only two had a solitary lesion, with others occurring at multiple locations, including the spine, hand, and pelvis.

Vaishya et al. reported a similar brown tumor at the tibia with the erosion of the posteromedial cortex, which was managed conservatively with below-knee cast immobilization for two months. The outcomes reported were tumor regression and clinical improvements, similarly at three months [5].

On the contrary, Naoya T reported a brown tumor of the right tibia managed with open reduction and internal fixation due to displacement. The patient was symptom-free at around two years after surgery [8]. Another case by Yalçin et al. was managed surgically by curettage and bone allograft insertion [6]. The patient was reportedly pain-free at around five months. The advantage is that the patient does not need to become exposed to the risk and complications of surgery.

The brown tumor may be in the differential diagnosis of solitary bone lesions in the young age group. Conservative management has a role in managing brown tumors of the lower extremities without complete fracture. Rapid diagnosis and removing the primary cause of parathyroid adenoma as soon as possible is a crucial part of the treatment.

Abbreviations

HPT: hyperparathyroidism; OPD: outpatient department

Conflict of interest

Nithi Pakmanee and Thanapon Chobpenthai declare that they have no conflict of interest.

Ethics approval and consent to participate

This study was approved by our institution's Human Research Ethics Committee (Chulabhorn Cancer Center, Bangkok, Thailand; project code: 177/2564). The study was performed following the ethical standards of the 1975 Declaration of Helsinki.

Availability of data and materials

Not applicable

Authors' contributions

All authors have read and approved the manuscript. NP: Conceptualization, Methodology, Validation, Formal analysis, Investigation, Data Curation, Writing - Original Draft, Visualization, Supervision, Project administration. TC: Validation, Formal analysis, Investigation, Writing - Original Draft, Supervision, Project administration, Writing - Review & Editing.

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