



Case Report

# Brown tumor due to primary hyperparathyroidism resulting in acute paraparesis: Case report and literature review

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## ABSTRACT

**Background:** Brown tumor (Osteoclastoma) is a rare benign, focal, lytic bone lesion most commonly attributed to a parathyroid adenoma; it occurs in approximately 5% of patients with primary hyperparathyroidism, and 13% of patients with secondary hyperparathyroidism. Most tumors are located in the mandible, pelvis, ribs, and large bones; only rarely is it found in the axial spine.

**Case Description:** A 37-year-old male with primary hyperparathyroidism presented with an MR-documented T4 and T5 brown tumor (Osteitis Fibrosa Cystica) resulting in an acute paraparesis. The patient successfully underwent excisional biopsy of an expansile, enhancing, bony destructive lesion at the T4-5 level. Subsequently, he required subtotal excision of a left upper parathyroid tumor.

**Conclusion:** Patient with primary hyperparathyroidism may acutely present with paraparesis attributed to brown tumors of the spine warranting emergent operative decompression.

**Keywords:** Osteitis fibrosa cystica, Primary hyperparathyroidism, Spine brown tumor

## INTRODUCTION

Brown tumor (Osteoclastoma) is a rare, benign, and focal lytic bone lesion that is most commonly attributed to a parathyroid adenoma. It occurs in 5% of primary and 13% of secondary cases of hyperparathyroidism.<sup>[8]</sup> These lesions are typically found in the mandible, pelvis, ribs, and large bones; only rarely do they involved the axial spine.<sup>[3]</sup> They are called brown tumors due to their high hemosiderin content. The term osteitis fibrosa cystica reflects their intrinsic peri trabecular bone fibrosis (osteitis fibrosa), and increased osteoclastic activity causing lytic bone lesions.<sup>[8]</sup> Notably, they may mimic metastatic lesions. Here, we present a 37-year-old male with an MR-documented T4-T5 thoracic brown tumor resulting in paraparesis due to primary hyperparathyroidism.

## CASE PRESENTATION

For 2 months, a 37-year-old male demonstrated a slow-growing lesion involving the dendro-alveolar segment of the left upper maxilla (rounded mass of 4 × 4 cm, nonulcerated with normal

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oral mucosa) [Figure 1]. The biopsy was consistent with a peripheral giant cell granuloma. Two months later, he newly presented with back pain, and 6 days of the lower extremity weakness. On examination, he exhibited bilateral iliopsoas/quadriceps (4/5)/bilateral extensor hallucis/dorsiflexor (2/5) weakness, and a left Babinski response. The laboratory studies revealed a marked elevation in both the calcium-corrected and alkaline phosphatase levels.

### Holospinal contrast enhanced MR

Multiple, expansile, enhancing, and bony destructive lesions were seen at many thoracic, lumbar, and sacral spinal levels (e.g., involving mainly the neural arches, and adjacent soft tissues). The largest lesions were at the T4/T5 level; the first contributed to marked central cord compression, while an additional very large, expansile, destructive bony lesion involved the left – T4-T5 ribs, and infiltrated into the adjacent paraspinal muscles. Other

similar and/or smaller lesions involved multiple spinous processes, lamina, and/or vertebral bodies (e.g., of C5-6, L2-L4, S1-S2, and the left iliac bone). There was also an accompanying dorsal thoracic scoliotic deformity convex to the right side [Figure 2].

### Surgery

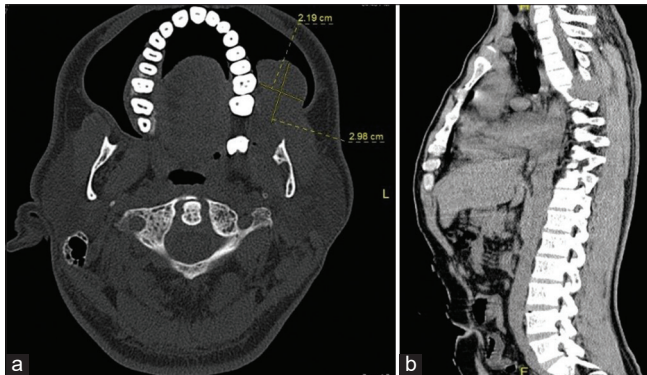
Due to significant cord compression, a T4/T5 coronal hemilaminectomy was performed with medial facetectomy/foraminotomy; care was taken to preserve the lateral two-third of the facet joints, and thus maintain stability. The soft and highly vascular T4-T5 tumor clearly compressed the thoracic cord, and infiltrated the para spinal ribs/muscle; tumor was readily debulked. The histopathology confirmed a brown tumor attributed to primary hyperparathyroidism [Figure 3]. Up to 2 years postoperatively, the patient remained neurologically intact with continued spinal stability.

### Diagnosis of primary hyperparathyroidism

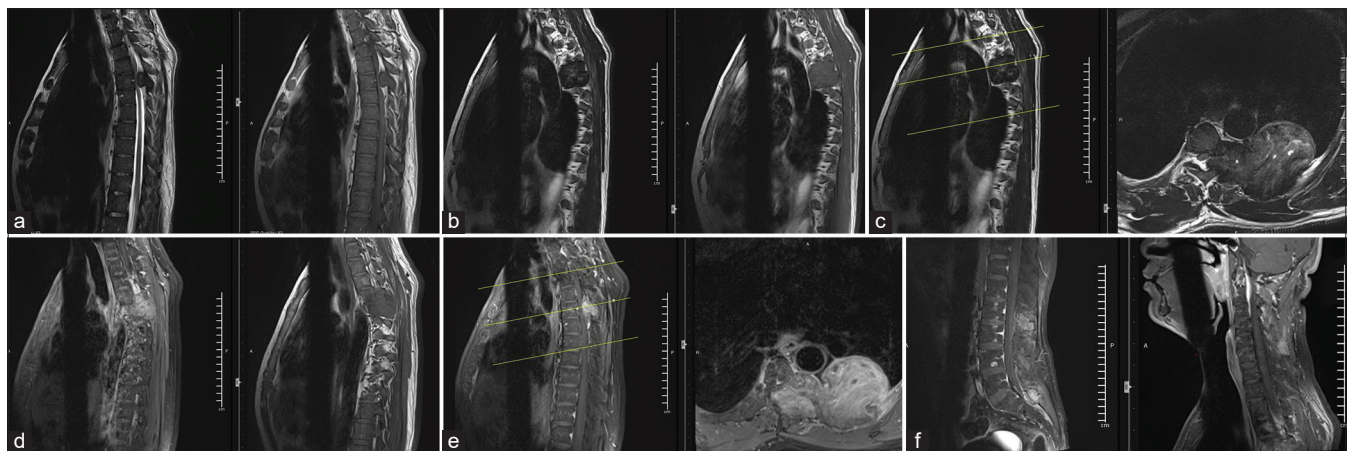
Primary hyperparathyroidism attributed to a parathyroid adenoma caused this patient's elevated parathyroid hormone (PTH) level (e.g., 1900 ph/ml). The patient subsequently required excision of the left upper parathyroid gland, following which calcium levels normalized [Figures 4-6].

### DISCUSSION

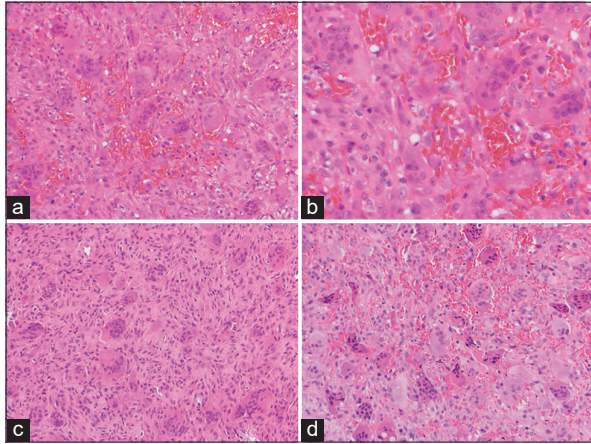
Brown tumors are rare benign, lytic bone lesions that appear grossly as soft, red brown expansile masses within the bone. They occur due to primary, secondary, or tertiary hyperparathyroidism.<sup>[6]</sup> Twenty-five similar cases of spinal brown tumors due to primary hyperparathyroidism have



**Figure 1:** (a) CT of maxillary sinus showing the left maxillary sinus mass. (b) CT thoracic spine is showing mass lesion in the upper thoracic area.



**Figure 2:** (a) MRI thoracic spine T2 sagittal on the left and T1 sagittal on the right. (b) MRI thoracic spine T2 sagittal on the left and T1 sagittal on the right (view more lateral to the left side). (c) MRI thoracic spine T2 sagittal and axial. (d) MRI thoracic spine T1 sagittal pre and post gadolinium-based contrast. (e) MRI thoracic spine T1 with contrast sagittal and axial. (f) MRI spine with contrast T1 with contrast lumbar spine sagittal on the left side and T1 with contrast sagittal cervical spine on the right side.

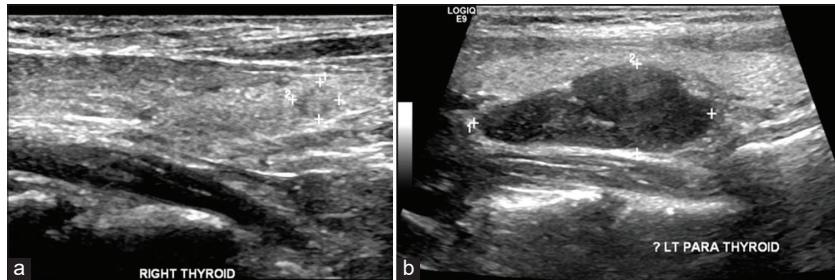


**Figure 3:** (a-d) Excisional biopsy shows cellular lesion composed of plump and spindle-shaped cells admixed with numerous multinucleated osteoclast-like giant cells with areas of hemorrhage and hemosiderin-laden macrophages. It was highly suggestive of brown tumor taking in consideration multiple bone involvement.

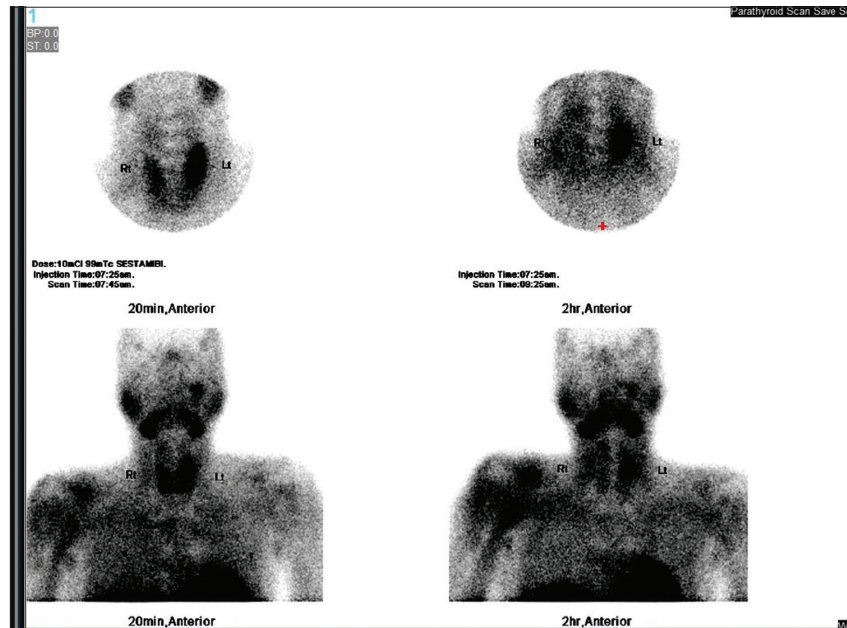
been described in the literature (e.g., 1968 to the present) [Table 1].<sup>[1-10]</sup> Patients typically range from 16 to 69 years of age, and more females than males are impacted (e.g., 15 females vs. 10 males). The most common location for brown tumors is the thoracic spine where presenting symptoms/signs typically range from back pain/radiculopathy, to myelopathy with paralysis with/without sphincter disturbances. Treatment options typically include: conservative management or surgery (e.g., biopsy/resection with/without instrumentation). Of the 25 prior cases reviewed, all except two underwent additional parathyroidectomy.

### Pathogenesis of brown tumors

Primary hyperparathyroidism and resultant spinal brown tumors are attributed to adenomas, resulting in elevated PTH, and heightened calcium levels; both contribute to abnormal bone mineralization. They also



**Figure 4:** (a) Thyroid ultrasound, right lobe of thyroid shows a small solid nodule measuring 4 × 5 mm noted at the lower aspect of the right lobe. Isthmus measures 2.5 mm. (b) Thyroid ultrasound, left lobe of thyroid at the region of the left parathyroid enlarged lobulated hypoechoic area noted measuring 34 × 13 mm with extremely increased vascularity, most likely representing a parathyroid adenoma.



**Figure 5:** NM parathyroid MIBI: Planar images of the neck in anterior projection were obtained 20 min and 2 h after IV administration of the radiotracer demonstrated, large persistent focal activity at the upper pole of the left thyroid lobe. No evidence of ectopic abnormal uptake.

**Table 1:** Reported cases of spinal brown tumor due to primary hyperparathyroidism.<sup>[4,7,8]</sup>

Author	Age/sex	Presentation	Location	Treatment spine	Parathyroidectomy
Shaw and Davies (1968)	58/F	Paraparesis urine retention	Thoracic	Resection	Yes
Shuangshoti <i>et al.</i> (1972)	32/M	Paraparesis radicular pain	Lumbar	Resection	Yes
Sundaram and Scholz (1977)	63/F	Paraparesis/urine retention	Thoracic	Resection	Yes
Siu <i>et al.</i> (1977)	64/F	Paraplegia/urine retention	Thoracic	Resection	Yes
Ganesh <i>et al.</i> (1981)	40/M	Paraparesis/radicular pain	Thoracic	Nil	Yes
Yokota <i>et al.</i> (1989)	58/F	Paraparesis/numbness	Thoracic	Resection	Yes
Daras <i>et al.</i> (1990)	54/F	Paraparesis	Thoracic	Resection	No
Kashkari <i>et al.</i> (1990)	51/F	Paraparesis	Thoracic	Resection + instrumentation	Yes
Sarda <i>et al.</i> (1993) <sup>1</sup>	23/F	Paraparesis, radicular pain	Thoracic	Resection	Yes
Motateanu <i>et al.</i> (1994)	57/M	Radiculopathy	Lumbar	Resection + instrumentation	Yes
Mustonen <i>et al.</i> (2004)	28/M	Radiculopathy, numbness	Lumbar	Nil	Yes
Haddad <i>et al.</i> (2007)	26/F	Paraparesis/numbness	thoracic	Resection	Yes
Khalil <i>et al.</i> (2007)	69/M	radiculopathy	thoracic	Resection	Nil
Altan <i>et al.</i> (2007)	44/F	Radiculopathy/low back pain	sacral	Resection	Yes
Hoshi <i>et al.</i> (2008)	23/F	Radicular pain	sacral	Nil	Yes
Lee <i>et al.</i> (2013)	65/M	Low back pain/radicular pain	lumbar	Resection + instrumentation	Yes
Khalatbari and Moharamzad (2014)	16/M	Paraparesis, sphincter dysfunction	lumbar	Resection + instrumentation	Yes
Khalatbari and Moharamzad (2014)	46/F	Paraparesis low back pain	lumbar	Resection	Yes
Khalatbari and Moharamzad (2014)	52/F	Radiculopathy, neck pain	cervical	Resection + instrumentation	Yes
Khalatbari and Moharamzad (2014)	38/M	Paraparesis, sphincter dysfunction	thoracic	Resection + instrumentation	Yes
Sonmez <i>et al.</i> (2015)	50/M	Paraparesis, sphincter dysfunction	thoracic	Resection + instrumentation	Yes
Alfawareh <i>et al.</i> (2015)	26/F	Radicular pain, quadriparesis	cervical	Nil	Yes
Hu <i>et al.</i> (2018)	50/F	Paraparesis	thoracic	Nil	Yes
Hammou <i>et al.</i> (2020)	65/F	Tetraparesis	cervical	Resection + instrumentation	Yes
Our present case	37/M	Paraparesis	thoracic	Resection mainly debulking	Yes

**Figure 6:** X-ray hand shows multiple osteolytic lesions.

result in decreased bone density, and increased bone resorption (e.g., due to heightened osteoclastic activity that progressively thins the bony cortex, and trabeculae). Resultant microfractures/microhemorrhages increase the activity of multinucleated macrophages, contributing to the formation of fibrous tissue resulting in osteitis fibrosa cystica. Notably, the typical brown tumors reflect the accumulation of hemosiderin.<sup>[6]</sup>

#### Differential diagnosis of brown tumors

The differential diagnoses for brown tumors include: metastases, multiple myeloma, sarcomas, giant cell reparative granulomas, lymphangiomatosis, leukemia, Langerhans cell histiocytosis, multiple bone cysts, aneurysmal bone cysts, and nonossifying fibromas.<sup>[2,6,8]</sup>

### Treatment of brown tumors: medical versus surgical management

There is an ongoing controversy regarding the comparative efficacy of parathyroidectomy with/without the need for attendant spine surgery.<sup>[4]</sup> Treatment of spinal brown tumors due to primary hyperparathyroidism typically includes initial resection of the parathyroid adenomas often normalizing PTH levels, contributing to remineralization, and the resorption of spinal lesions.<sup>[4]</sup> However, for patients presenting with significant spinal cord compression due to brown tumors, immediate operative decompression/stabilization may be necessary, followed secondarily by resection of parathyroid adenomas.<sup>[1,3,5,8,9]</sup> For the case presented, the patient's initial significant paraparesis due to the T4-T5 largest compressive spinal lesions warranted operative decompression, both to reverse the neurological deficit/paraparesis, and provide a tissue-diagnosis.

### CONCLUSION

Patients with primary hyperparathyroidism may acutely present with paraparesis attributed to thoracic brown tumors warranting emergent operative decompression, followed by resection of parathyroid adenomas.

### Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

### Financial support and sponsorship

Nil.

### Conflicts of interest

There are no conflicts of interest.

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