Saudi Journal of Radiology
6 2022 1(1), 28–35
Case Report

SPINAL CORD COMPRESSION AS THE FIRST PRESENTATION OF POLYOSTOTIC BROWN TUMOR: A CASE REPORT AND REVIEW OF IMAGING FEATURES

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ABSTRACT

Despite the fact that brown tumor is the hallmark of hyperparathyroidism, it is rarely observed due to early laboratory detection of elevated parathyroid hormone levels. The unusual occurrence of this entity in everyday practice is a concern not only for patients but also for physicians, as it can be mistaken for other lytic bone lesions. A diagnosis can easily be made if the full continuum of clinical, laboratory and radiological findings is considered. Herein, we report a case of polyostotic brown tumor presenting with thoracic cord compression secondary to vertebral pathological fracture.

الملخص

يعتبر الورم البني في العظم أحد أبرز الأورام الناتجة عن إرتفاع هرمون الغدرة الجار درقية, علماً بأنه أصبح نادر الوجود في وقتنا الحالي بسبب التشخيص المبكر لارتفاع مستوى هرمون الغدة الجار درقية في الجسم. ندرة وجود هذا النوع من الورم خلق نوع من القلق للمرضى والأطباء حيث أنه غالباً ما يتم تشخيصه بأنواع أخرى من أورام العظم الأكثر انتشاراً والتي قد تكون خبيثة. يصبح تشخيص الورم البني في العظم سهلاً اذا تم أخذ جميع المعلومات الإكلينيكية للمريض والتحاليل الطبية والأشعة التشخيصية بعين الاعتبار. ننشر في هذا المقال حالة مرضية لشخص مصاب بالورم البني في عظام متفرقة في الجسم والتي نتجت عن كسر في العمود الفقري وإصابة في الحبل الشوكي.

Received 24 May 2022
Revised 12 June 2022
Accepted 23 June 2022
Published 27 June 2022

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eISSN: 1658-8959

Keywords: Chronic kidney disease, Brown tumor, Hyperparathyroidism, Spinal cord compression, Pathological fracture

1. INTRODUCTION

Brown tumor has been widely reported in the medical literature with greatly variable symptomatic and background presentations. It is considered a benign focal reactive, slowly growing bone lesion seen in 3% of patients with primary hyperparathyroidism and 1.5%-1.7% of patients with secondary hyperparathyroidism [1].

The incidence of brown tumors is extremely rare in developed countries because hyperparathyroidism is sufficiently early detected using routine laboratory and radiological investigations. Clinically, patients with brown tumor may present with bone pain, pathological fracture or slowly growing palpable bone swelling. The pain experienced from the brown tumor is comparatively milder than that of malignant bone lesions [1]. As a result, the brown tumor is often found incidentally on imaging modalities requested for another indication [2]. In such cases, a dedicated computed tomography (CT) or magnetic resonance imaging (MRI) is often requested to characterise the lesion further and assess the full extent of musculoskeletal involvement.

Case Report

A 45-year-old male known case of end-stage renal disease (ESRD) on hemodialysis presented to the emergency department with a one-day history of severe mid to lower back pain associated with the inability to walk and numbness from the thorax to the toes bilaterally. There was no sphincter dysfunction. The power was five out of five bilaterally on physical examination, with grade two reflexes. In addition, the patient had para-vertebral tenderness at the thoracic and lumbar spine.

A whole spine MRI was done to exclude spinal cord compression and showed a compression fracture of the T6 vertebra with retropulsion of the posterior vertebral border, causing narrowing of the spinal canal and spinal cord compression (Figure 1).

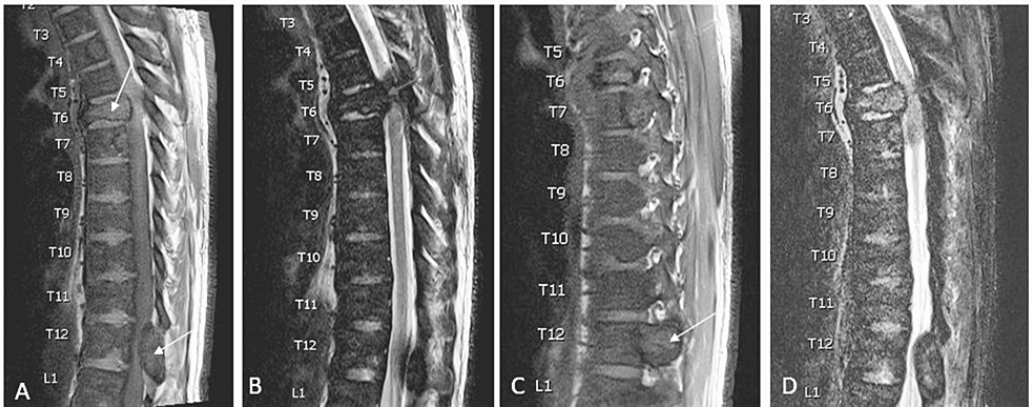

Figure 1 Sagittal T1 (A), T2 (B and C) and STAIR (D) MRI sequence of the thoracic spine showing diffuse abnormal bone marrow signal intensity with multilevel expansile osseous lesions involving the anterior and posterior column of the spine (white arrows). A pathological compression fracture of the T6 vertebra involving 40% of vertebral height is associated with retropulsion and contributes to spinal canal narrowing (red arrow in B). At the same level, the surrounding intraspinal CSF space is complete effacement associated with abnormal high T2 and STIR signal intensity within the spinal cord (red arrow in D).

In addition, MRI showed a right posterior fifth rib lesion partially visualized. For this reason, a contrast-enhanced CT scan of the chest, abdomen and pelvis was requested for the patient to evaluate the extent of the osseous lesions further and to look for primary

malignancy; as the presence of multiple osseous lesions and pathological compression fracture raised the possibility of osseous metastasis as one of the differential diagnosis. CT scan of the abdomen and pelvis showed multiple vertebral and rib lesions with no obvious primary malignancy (Figure 2A-C). Furthermore, the kidneys are atrophied with multiple small, simple cortical cysts (Figure 2D). These findings support the patient's known history of chronic kidney disease.

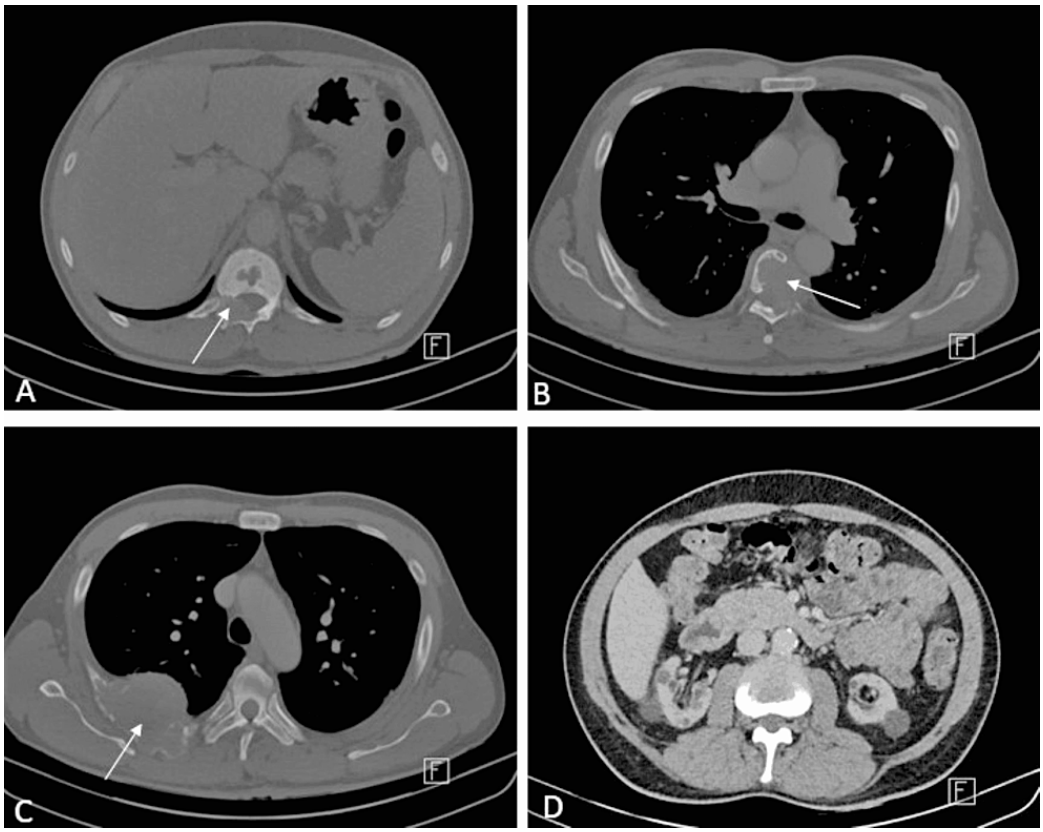

Figure 2 Axial CT scan of the chest and abdomen. Expansile lytic lesion at T12 vertebra (A) and T6 vertebra extending to the right pedicle and lamina (B). Large destructive expansile lytic lesion involving the right posterior fifth rib (c). The kidneys are atrophic with multiple bilateral simple cysts (D).

The patient underwent thoracic spine decompression and fixation with uneventful surgery. Post-operative chest radiograph is shown (Figure 3).

During his hospital stay, the patient experienced left-hand pain for which a radiograph showed an expansile lytic lesion (Figure 4).

After excluding metastasis as a differential diagnosis of such multiple lytic lesions, the brown tumor was considered a possible differential considering the patient's history of ESRD. Serum calcium and parathyroid hormone level was requested and showed low calcium level measuring 1.9 mg/dL and elevated parathyroid hormone measuring 471 picograms per millilitre (pg/mL), suggestive of secondary hyperparathyroidism. The

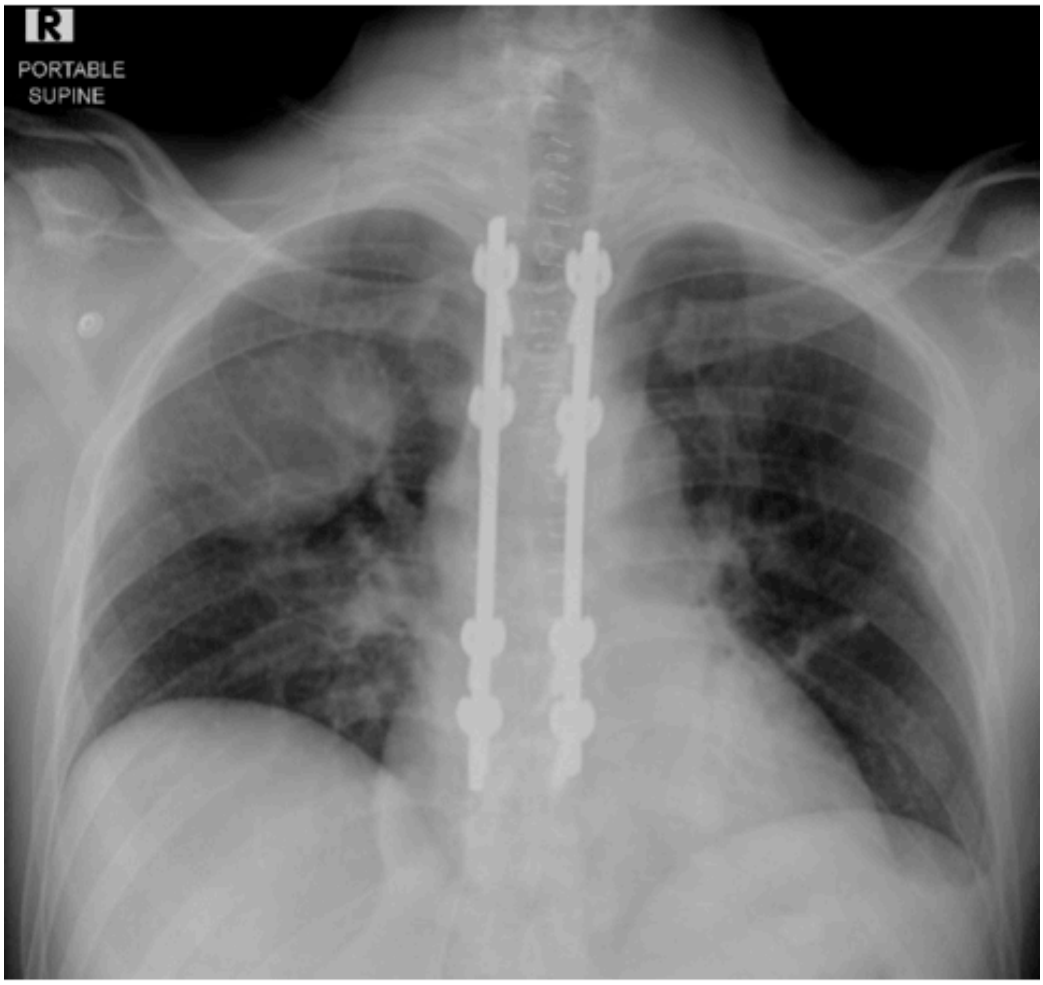

Figure 3 Frontal chest radiograph post spinal surgery. Thoracic spinal fixation hardware, surgical clips, and drain in situ—bilateral expansile rib lesions of variable size.

parathyroid nuclear scan was requested to complete the workup, which confirmed the diagnosis of parathyroid gland hyperplasia (Figure 5 and 6). The patient was started on oral calcimimetics and remarkably improved diffuse bone pain.

Figure 4 Frontal (A), lateral (B) and oblique (C) radiograph of the left hand. A well-defined expansile lytic lesion with multiple internal septations and cortical irregularity along the lateral aspect at the head of the left third metacarpal bone. Another small lytic lesion at the distal, proximal phalanx of the thumb.

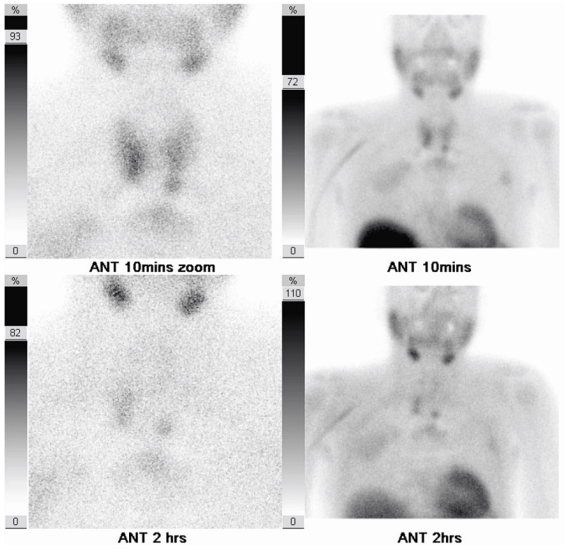

Figure 5 Parathyroid nuclear scan using Technetium (99mTc) sestamibi. Anterior (ANT) projection images acquired at 10 minutes show asymmetrical radiotracer uptake in both thyroid lobes, extending down on the right lower side. Anterior (ANT) projection images acquired at 2 hours show retention of radiotracer in bilateral lower thyroid bed with uniform washout of radiotracer from rest of the thyroid bed.

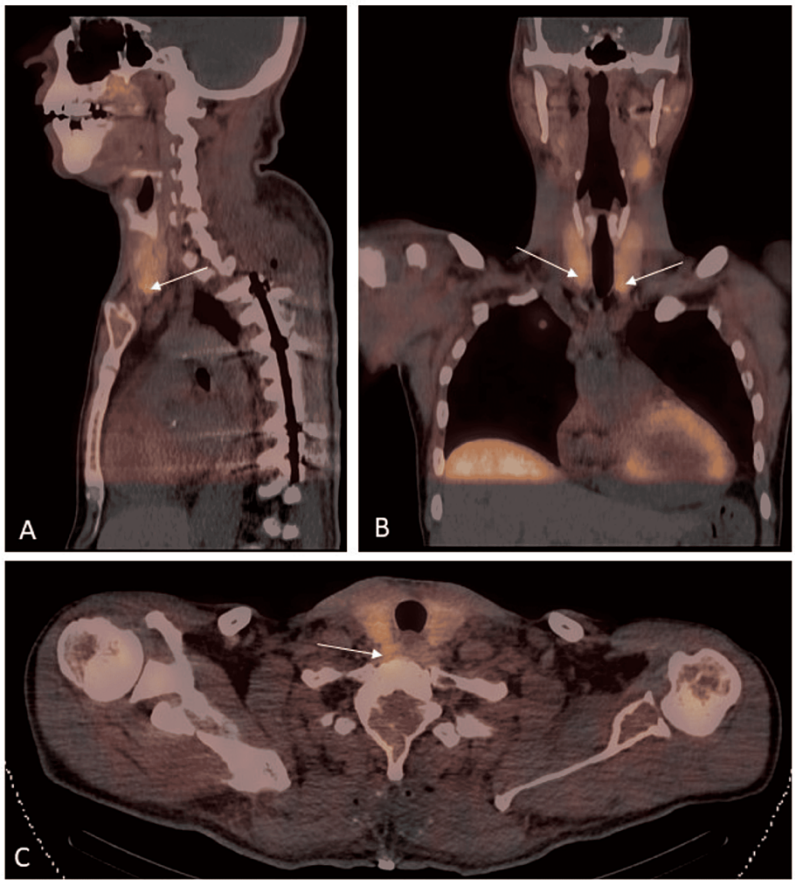

Figure 6 Sagittal (A), coronal (B) and axial (C) SPECT-CT images at 20 minutes. Focal increased uptake at the lower aspect of the right and left thyroid lobes (white arrows) suggests hyperplasia of bilateral lower parathyroid glands.

2. DISCUSSION

This case report is of special interest because of the multiplicity and imaging feature variability of a single osseous pathology. Interestingly, the brown tumor is usually seen in the end spectrum of renal osteodystrophy, which is associated with secondary hyperparathyroidism [1, 3]. However, in this case, the sole presence of brown tumors and the absence of other radiological features of renal osteodystrophy makes the case distinctive and teachable value.

In hyperparathyroidism, osteoclasts' increased activity results in osteopenia and subperiosteal bone resorption. At the same time, the resultant reactive osteoblastic activity leads to the ingrowth of fibrovascular tissue in the bone marrow. Collectively, this results in microfractures and haemorrhages within the marrow cavity-causing influx of multinucleated macrophages [4]. The presence of hemosiderin within the tissues gives the lesion a brownish appearance in gross pathology, hence its name [5].

Brown tumors can be monostotic or polyostotic [1]. Typically, it arises in the hands, pelvis, ribs, facial bones, and femur [3, 6, 7]. Spine involvement of brown tumor is relatively rare, with only 18 cases have been reported in the literature [2]. CT appearance of brown tumor is non-specific and can mimic other malignant bone lesions like metastases, multiple myeloma or benign cystic bone lesions. It may have well-defined sclerotic margins or be expansile with cortical destruction and soft-tissue mass, suggesting an aggressive bone tumor. In fact, the majority resemble bone cysts or other benign lesions. [1, 6, 8]. In MRI, the presence of hemosiderin in the brown tumor and the lytic appearance make it resemble other hemosiderin-containing benign lesions like giant cell tumors, giant cell reparative granulomas, hemorrhagic bone cysts, bone infarcts, pigmented villonodular synovitis or hemophilic pseudotumors. Due to magnetic susceptibility, hemosiderin within the tissues results in a signal loss in gradient echo images [5].

3. CONCLUSION

Brown tumor is a rare bony lesion resulting from hyperparathyroidism. Besides biochemical investigation for hyperparathyroidism, sestamibi scintigraphy should be performed to localize the active parathyroid gland. A patient with a brown tumor may present with insidious pain, localized bone swelling or pathological fracture like in this case. Brown tumor exhibits various radiological features, including single or multiple lytic lesions. In addition to conventional radiology, cross-sectional imaging should be performed to distinguish it from other bone tumors that should be considered even in the background of hyperparathyroidism.

CONFLICT OF INTEREST

The authors declare that the research was conducted without any commercial or financial relationships that could be construed as a potential conflict of interest.

ACKNOWLEDGEMENTS

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