

## Brown tumour as an unusual but preventable cause of spinal cord compression: Case report and literature review

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### Abstract

Brown tumour is an infrequent, focal, and benign osteolytic lesion which is a consequence of abnormal bone metabolism in hyperparathyroidism (both primary and secondary). It is also known as Osteoclastoma. In the present era, we rarely encounter skeletal disease caused by primary hyperparathyroidism. Although it is a rare presentation because of advancement of treatment but still can be encountered because of lack of standard care so we should have high index of suspicion to avoid this preventable complication. We report here a case of brown tumour in the thoracic vertebra of a young female patient with End Stage Renal Disease, who presented with backache and bilateral lower limb weakness. MRI of the spine showed multiple non-enhancing abnormal signals involving vertebral body of C2, posterior elements of C6, and bilateral sacral vertebra, suggestive of healed fractures versus bone forming tumours. She underwent laminectomy. Her histopathology report was consistent with brown tumour of hyperparathyroidism.

**Keywords:** Hyperparathyroidism, brown tumour, laminectomy, osteoclastoma, end stage renal disease.

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### Introduction

Hyperparathyroidism (HPT) results in an augmented efflux of calcium from the bones due to which plasma calcium's concentration is increased, which leads to reduced absorption of calcium by the kidneys and intestine resulting in skeletal demineralisation; hence, the bone is being replaced by osteoclasts (or multinucleated giant cells) which contribute to deformed and labile skeleton.

Primary hyperparathyroidism is caused by hyper secretion of the parathyroid hormone due to changes in one or more parathyroid glands. The end result is hypercalcaemia. Secondary hyperparathyroidism is the result of end-stage renal disease (ESRD) in which there is excessive loss of calcium in the urine, which leads to elevation of serum

parathyroid hormone level secondary to feedback response to low serum calcium levels. Tertiary hyperparathyroidism is seen in patients who have longstanding chronic kidney disease. In tertiary hyperparathyroidism there is autonomous hyperplasia of the parathyroid glands as there is no feedback response to the level of calcium in serum. The pathognomic feature of hyperparathyroidism is resorption of skeleton which occurs due to augmented activity of osteoclasts. There is accumulation of loose connective tissue inside both cortical and trabecular bones. In some patients, brown tumour of hyperparathyroidism is seen which is a distinct lesion composed of osteoclasts, numerous reactive giant cells and haemorrhagic debris.<sup>1</sup>

In end stage renal disease, a variety of bony involvement is seen which is termed as renal osteodystrophy. In our case, HPT was diagnosed by the presence of osteolytic lesions called osteoclastomas or brown tumours. Only a few patients with HPT have radiologically noticeable skeletal changes. The characteristic skeletal manifestations of HPT are mentioned below:

- (1) Erosions of periosteal surfaces of phalanges of the hands.
- (2) Ten percent of HPT cases have brown tumours and these usually appear in advanced disease.
- (3) There are localised regions of bone loss due to augmented osteoclastic activity which leads to loss of architecture of the skeleton; these are termed as Osteitis fibrosa cystica.
- (4) The skeleton has radiolucent appearance which is secondary to demineralisation.
- (5) Due to loss of central trabeculae and thinning of the cortex, the skull bones have granular appearance in HPT.<sup>2</sup>
- (6) Pathologic calcifications with punctuate appearance are seen in soft tissues, kidneys and joints.

In this case report, we present a young female patient with brown tumour of thoracic spine causing spinal cord compression so as to raise its awareness among health care professionals since this is rare but preventable cause of

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spinal cord compression. The possibility of brown tumour must be kept in mind in the differential diagnosis of sudden neurologic impairments and paraplegia/paraparesis in chronic kidney disease patients with secondary HPT.

### Case Report

A 42-year-old female, a known case of hypertension and end stage renal disease (for eight years) and history of parathyroidectomy (in 2017), now presented in Aga Khan University Hospital Karachi, in emergency department in June 2019 with diffuse generalised bone pains, lower back pain, and bilateral lower limb weakness. Examination showed right lower limb power of 4/5 and left lower limb power of proximal of 3/5 and distal of 4/5. MRI of the spine showed multiple non-enhancing abnormal signals involving vertebral body of C2, posterior elements of C6, and bilateral sacral vertebra, suggestive of healed fractures versus bone forming tumours. Multiple enhancing lesions involving the D9 vertebrae, ribs, skull base, and jaw were also observed. There was collapse of D9 with retropulsion of the body, resulting in cord compression. [Figure]. Blood investigations showed calcium of 9.6 mg/dl (NR 8.6-10.2 mg



**Figure:** MRI spine: An enhancing lesion involving D9 vertebrae. There is collapse of D9 with retropulsion of the body.

**Table:** Investigations.

Investigations	Results	Normal ranges
<b>Pre-operative</b>		
Calcium	9.6	8.6-10.2 mg/dl
Albumin	3.2	3.5-5.2 g/dl
Phosphorus	4.8	2.5-4.5 mg/dl
25 OH Vitamin D	9.96	>30 ng/ml
PTH levels	744	16-87 pg/ml
<b>Post-operative</b>		
Calcium	6.2	8.6-10.2 mg/dl
Albumin	3.2	3.5-5.2 g/dl
Phosphorus	4.1	2.5-4.5 mg/dl
25 OH Vitamin D	32	>30 ng/ml
PTH levels	39.8	16-87 pg/ml

/dl) with albumin 3.2, Phosphorus 4.8 mg/dl (NR 2.5-4.5 mg/dl), Vitamin D level of 9.96 ng/ml (NR >30 ng/ml), and PTH 744 pg/ml (NR 16-87 pg/ml). [Table] Due to significantly raised PTH level the patient underwent Sestamibi scan which turned out to be normal.

Due to cord compression, our patient underwent T9 laminectomy. Histopathology showed multiple light brown tissue fragments measuring 3x2x1 cm in size, it was a giant cell rich lesion, and the features were suggestive of brown tumour of hyperparathyroidism.

The patient was given injection Vitamin D 200,000 IU (one dose only) and Cinacalcet was started. After two months during clinic follow-up visit her PTH was noted to have risen to 2314 ng/ml. Sestamibi scan was repeated, which turned out to be normal. Ultrasound of the neck revealed hypo echoic lesions at the dorsal lower poles of both the thyroid lobes; these findings were consistent with parathyroid adenomas. Her Cinacalcet was continued (she was advised compliance in its use). She finally underwent parathyroidectomy. Her postoperative PTH was 39.8 pg/ml (NR 16-87 pg/ml), and Vitamin D of 32 ng/ml (NR >30 ng/ml). She developed hungry bone syndrome (with postoperative calcium of 6.2 mg/dl); IV Calcium gluconate 2gm six hourly, Qalsan D (Chewcal) 4 tabs every six hour and Alphacalcidol (activated form of Vitamin D) 1mcg BID were started. Her calcium level was frequently monitored in special care unit. Gradually, her calcium level improved and she got better.

### Discussion

In the presented case, the features in favour of the diagnosis of brown tumour are hyperparathyroidism, the presence of giant cells in the spinal histopathology, and End stage renal disease. Hungry bone syndrome (HBS) is very rapid, robust, severe, and prolonged hypocalcaemia and is worsened by low parathyroid hormone (PTH) levels. Hungry bone syndrome is associated with hypophosphataemia and hypomagnesaemia. It is seen mostly in patients who have had parathyroidectomy, especially in those patients who had preoperative high bone turnover and severe primary hyperparathyroidism (PHPT). On histopathology, brown tumour specimen of hyperparathyroidism contains spindle cell proliferation, large amount of haemosiderin and multinucleated giant cells, and it looks like giant cell tumour.<sup>3,4</sup> Due to haemorrhage in it and haemosiderin accumulation, it is dark, reddish-brown in colour. These bone-resorbing lesions can occur in any part of the skeleton. Brown tumours gradually disappear after parathyroidectomy.<sup>5</sup> These tumours are rarely encountered and are seen only in 3% of primary hyperparathyroidism and 1.5% in secondary

hyperparathyroidism.<sup>6,7</sup> The common sites where brown tumour is seen are sternum, ribs, phalanx, mandible, maxilla, pelvic bones, and femur. These can arise as solitary or multiple lesions.<sup>8</sup> Osteitis fibrosa cystica or brown tumours originate because of augmented osteoclastic activity and fibrosis of peritrabecular areas which occurs secondary to phosphate retention and defective synthesis of Vitamin D.<sup>9</sup> This tumour is a peculiar feature of both primary and secondary hyperparathyroidism. Intraspinous and paravertebral soft tissue masses along with bone infiltration and neural compression are detected on magnetic resonance imaging in patients with brown tumour of the spine. Brown tumour appears hyper/hypo intense on T2-weighted images and hypo intense on T1-weighted images on MRI.<sup>10</sup>

### Conclusion

Brown tumour is an unusual manifestation of hyperparathyroidism, it is more common with secondary hyperparathyroidism than primary hyperparathyroidism. Our case is unique in the context of site of involvement because brown tumour rarely involves spine. Brown tumour should be considered in the aetiology of spinal lesions in patients with end stage renal disease. The physician should be aware of the clinical manifestations and radiographic appearances of brown tumours to allow for timely diagnosis, treatment and improvement of prognosis of the disease. In addition to plain X-ray, CT and/or MRI of spine should be performed to help differentiate it from other skeletal tumours. If there is any doubt in the diagnosis, biopsy of the bone tumour is recommended for definitive histopathological diagnosis.

**Consent:** The patient provided a written consent for publishing her case report for enhancement of science.

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