

## A CASE REPORT OF FLORID CLASSICAL PRESENTATION OF SOLITARY PARATHYROID ADENOMA WITH BROWN'S TUMOUR AND HUNGRY BONE SYNDROME

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### ABSTRACT

#### BACKGROUND

A 19 years old young female was admitted to our Tertiary Care Referral Institute as a case of solitary parathyroid adenoma with hyperparathyroidism and Brown's tumour. The serum calcium and PTH levels were significantly raised, ultrasonography and CECT neck was suggestive of a nodule at the lower pole of left lobe of thyroid, supplied by inferior thyroid artery, possibly parathyroid adenoma. FNAC of swelling on the left hard palate was suggestive of Brown's tumour. Both the tumours were surgically removed and confirmed with histopathological examination as parathyroid adenoma and Brown's tumour respectively. Post surgery hungry bone syndrome was managed successfully.

#### KEYWORDS

Parathyroid Adenoma, Brown's Tumour, Parathyroidectomy, Hungry Bone Syndrome.

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#### BACKGROUND

A solitary parathyroid adenoma is the most common cause of primary hyperparathyroidism. It is characterised by elevated serum calcium levels with unsuppressed parathyroid hormone levels. Primary hyperparathyroidism is usually seen in females above the age of 50 years with a prevalence of 21/1000<sup>1</sup>, whereas the incidence in patients aged 12 - 28 years is less than 5%.<sup>2</sup> The sex ratio is 1:3 (male-to-female). The primary hyperthyroidism is of two types, sporadic (95%) and hereditary (5%); 80% of sporadic cases are due to solitary adenoma,<sup>3</sup> 15 - 20% due to glandular hyperplasia and 1% are due to carcinoma. Hereditary pHPT is associated with MEN 1 syndrome.

In primary HPT, about 50% of patients have no symptoms and the problem is picked up as an incidental finding (via raised calcium or characteristic x-ray appearances [subperiosteal resorption of the phalanges of the index and middle fingers]).<sup>4</sup> Most of the patients present with non-specific symptoms of fatigue, lethargy, depression, loss of concentration, bone and joint pain.

Very few patients present with the classical rhyme of bone, stones, abdominal groans and psychiatric moans as described in 1930's, which is implied as a late presentation. The incidence of hyperparathyroidism associated with a Brown's tumour is less than 5%.<sup>5</sup> It is important to evaluate for the causes of hyperparathyroidism esp. MEN 1 in young adults. To evaluate the patient of solitary parathyroid adenoma non-invasive studies include ultrasonography, sestamibi scan, CECT and MRI.

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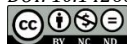
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Invasive procedure include image guided Fine Needle Aspiration Cytology and the PTH assay. The treatment of choice is parathyroidectomy.

#### Case Summary

A 19 years old young female, resident of Shimla, was referred to Department of ENT at Indira Gandhi Medical College, Shimla, India on 15 May 2016 with complaints of off and on pain in the right flank and right hip for the last two months, which was insidious in onset and gradually progressive in nature. It was associated with easy fatigability, vague abdominal pain, off and on nausea, generalised weakness, body aches, mood swings and pain in the right upper jaw. There was no history of increased frequency of urination, increased thirst or fractures.

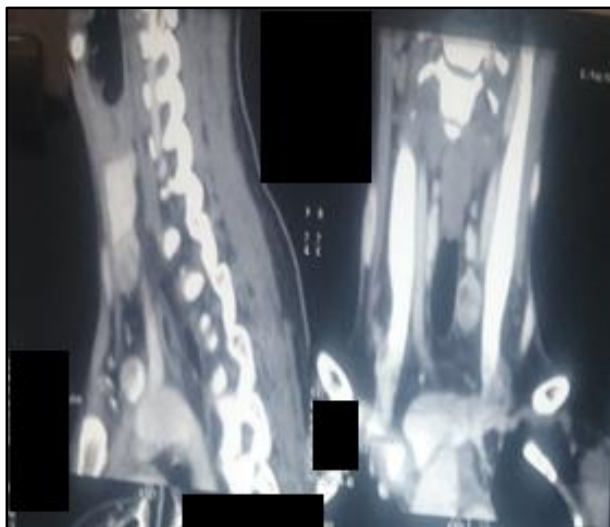
On systemic examination of per abdomen tenderness was present in the right iliac fossa with no signs of guarding and rigidity or any appreciable organomegaly. On examination of oral cavity, a 4 × 3 cms firm and tender swelling present on the left side of hard palate and examination of neck was within normal limits. On locomotor examination, limp gait was present with tenderness on the right hip and normal sensory system examination.

The patient was investigated, the serum calcium levels were 16.1 mg/dL, serum PTH was 1607.8 pg/mL and 24 hours urine calcium was 191.1 mg/24 hours.

Doppler ultrasonography of neck was suggestive of a hypoechoic nodule of size of 1.9 × 1.0 cms at the lower pole of left lobe of thyroid, supplied by inferior thyroid artery, possibility of parathyroid adenoma.

USG abdomen showed multiple echogenic foci, maximum of 6 mm floating in the gall bladder with wall thickness of 4 cms, suggestive of calculus. Right kidney was normal in size with echogenic focus of 1.2 cms in the middle calyx, suggestive of calculus.

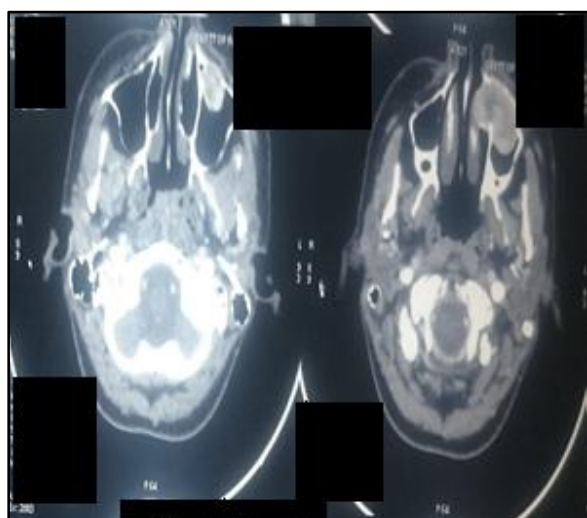
CECT neck showed a well-defined nodule of size of 15 × 13.4 mm with craniocaudal extent of 2.1 mm, seen in relation with the inferior lobe of left thyroid gland.



**Figure.1: CECT Neck- nodule of size of 15×13.4mm with craniocaudal extent of 2.1mm, suggestive of parathyroid adenoma**



**Figure.2: X-ray PNS- Hazy left maxillary sinus**



**Figure.3: HRCT of paranasal sinuses showed a radio-dense lesion in the left maxilla, extending to the left maxillary sinus**



**Figure.4: X- ray forearm showed osteoporosis**

The Bone Scan of patient showed a T-SCORE of 3.2, which was consistent with the findings of osteoporosis.

FNAC of hard palate swelling was suggestive of multinucleated giant cells of osteoclast type with occasional osteoblast, suggestive of Brown’s cell tumour.

Patient then underwent excision of the parathyroid adenoma under general anaesthesia, per operatively an adenoma of size of about 1.5 × 1.5 cms with smooth surface, firm consistency, lying in relation to inferior thyroid artery was excised in total and sent for histopathological examination, which was suggestive of parathyroid adenoma.



**Figure.5. parathyroid adenoma in relation to inferior thyroid artery**

On post-operative day one, serum PTH levels and serum calcium levels were 87.1 pg/mL and 7.2 mg/dL respectively.

Then patient went in hungry bone syndrome with serum PTH and serum calcium levels as 873 pg/mL and 7.4 mg/dL respectively, which was successfully managed with calcium and vitamin D supplements.

After one month, patient underwent excision of Brown’s tumour of left maxilla through Caldwell-Luc approach under general anaesthesia. Per operatively, a red coloured mass of size of 4×4 cms was curetted out in total and sent for histopathology, features of which were consistent with Brown’s tumour.

The recent serum calcium and PTH levels were 8.8 mg/dL and 60 pg/dL respectively. The patient has symptomatically improved with no limp.

#### DISCUSSION

Solitary parathyroid adenoma is associated with inappropriate increased secretion of PTH, which affects the serum calcium and phosphate levels. Thereby, it affects the bone metabolism due to hypercalcaemia and hypophosphataemia. However, the clinical manifestations of hyperparathyroidism are variable, about 80% are asymptomatic and 20% to 30% present with recurrent nephrolithiasis, osteoporosis, proximal muscle weakness and psychiatric symptoms and Brown's tumour.

Brown's tumour also known as osteitis fibrosa, cystica generalisata or Von Recklinghausen's disease of bone, is a metabolic bone disease that develops in primary, secondary or tertiary HPT. It represents reparative granuloma rather than a true neoplastic process.<sup>6,7</sup> It is a benign bony lesion caused by localised osteoclastic turnover of bone under the effect of increased PTH with replacement of fibrous tissue and giant cells. Brown's tumour are one of the most pathognomonic sign of pHPT. They can be located in any part of the skeleton, but are most frequently encountered in the ribs, clavicles, extremities and pelvic girdle. Clinically, significant lesions in the craniofacial bones are rare.<sup>8</sup> Furthermore, the frequency of occurrence is more among persons older than 50 years of age with a male-to-female ratio of 1:3.<sup>9,10</sup>

Our case is unique, as almost all the pathogenic signs of primary hyperthyroidism are present in a single case at a young age. The patient had nephrolithiasis, cholelithiasis and osteoporosis, Brown's tumour of left maxilla, psychiatric symptoms in the form of mood swings and post-operative hungry bone syndrome which was managed successfully with calcium and vitamin D supplements. HBS is currently considered actual till postoperative hypocalcaemia recovers after parathyroidectomy and calcium supplementation can be safely interrupted.<sup>11</sup> This occurs when bone turnover turns back to normal and the function of the residual non-pathogenic glands regains normal PTH secretion levels.

The patient is in regular followup with latest serum PTH and calcium levels as 60 pg/mL and 8.8 mg/dL respectively. Nephrolithiasis, cholelithiasis and osteoporosis was successfully managed by Surgery and Orthopaedic Department.

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