

## Case Report

# A large parathyroid adenoma presenting with pathological fractures in a young male

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### ABSTRACT

Parathyroid adenoma is the common cause of primary hyperparathyroidism and usually presents with features of hypercalcaemia. In the present case report, we describe a case of a large adenoma of the parathyroid gland presenting with pathological fractures. A 42 year old Male presented with bony pains and fracture of the left humerus after a trivial trauma. A detailed clinical examination and laboratory studies revealed hypercalcaemia secondary to hyperparathyroidism. The patient underwent targeted parathyroidectomy of the involved gland after appropriate preoperative localization. Histopathology of the resected specimen was consistent with parathyroid adenoma. Conclusion is a common cause of primary hyperparathyroidism is a common endocrine disorder occurring as a result of Parathyroid adenoma in 80-85% cases. Patients usually present with symptoms related to hypercalcaemia. A high index of suspicion, careful preoperative workup, localization of the affected gland(s) and surgical excision ensures a successful cure.

**Keywords:** Parathyroid adenoma, Hyperparathyroidism, Hypercalcaemia, Sestamibi

### INTRODUCTION

Hyperparathyroidism usually presents with features of hypercalcaemia such as abdominal pain, fatigue, bony pains, nephrolithiasis and neuropsychiatric symptoms. It is classified into three main groups. Primary hyperparathyroidism is due to parathyroid adenomas in 80-85% cases, gland hyperplasia in 10-15% and rarely carcinoma in <1% cases.<sup>1</sup> Parathyroid adenoma is a benign neoplasm commonly involving single gland, but can also involve multiple glands also.<sup>2</sup> Patients presenting with osteoporosis is a rare entity. Pathological fractures seen in severe hyperparathyroidism can be associated with classical skeletal complication of hyperparathyroidism called osteitis fibrosa cystica, although it is very rare in the modern era and occurs in approximately 2% of patients diagnosed with

hyperparathyroidism.<sup>3</sup> It is associated with loss of bony mass in the matrix due to increased osteoclastic activity in hyperparathyroidism. The resultant weakening of the bones because of replacement of the calcified supporting structures with fibrous tissue, there is formation of cyst like brown tumors in the bone resulting in increased risk of fractures. There are number of studies which have confirmed that there is increased risk of fractures in patients with hyperparathyroidism and a reduced risk in patients who undergo parathyroidectomy.<sup>4</sup> Surgical excision of the hypersecretory gland is the mainstay in management which may be traditional neck exploration or minimally invasive parathyroidectomy. Use of preoperative localization techniques using Ultrasonography (USG), Computed Tomography (CT) scan and nuclear imaging or intraoperative parathormone assay to ensure resection of the target hypersecretory

gland are important adjuncts in aiding the surgeon. Technetium Tc99m sestamibi is the radio tracer of choice, with sensitivity reported to be between 72-100%.<sup>5</sup> There is a growing trend towards minimally invasive surgery with adequate preoperative localization and intraoperative parathormone monitoring.

**CASE REPORT**

A 42 year old male non-smoker without any known comorbidities presented with history of bony pains, generalized lassitude, constipation and non-healing fracture of left humerus sustained three months ago with trivial trauma while lifting some weight. Patient also gave history of fracture left forearm which was managed with open reduction and internal fixation three years ago. There was history of renal calculi two years back which was managed conservatively. There was no other significant family or personal history. Radiographs revealed fracture of left humerus and fracture of left ulna with K-wire in situ. Investigations (Table 1) revealed hypercalcaemia, elevated alkaline phosphatase, decreased vitamin D3 levels, elevated parathormone, normal phosphate levels and renal parameters.

There was evidence of generalized osteoporosis on radiographs of the small hand bones, pelvis, skull and spine and also on bone densitometry (DEXA scan). Whole body bone scan showed patchy tracer uptake in entire skeleton with generalized osteoporosis suggestive of metabolic bone disease. USG of the neck revealed an oval hypoechoic lesion measuring 1.9 x 2.5 x 4.7 cm noted on the posterior aspect of the right lobe of thyroid gland with few calcific foci within and peripheral vascularity.

A contrast enhanced CT of the neck showed a moderately large oval heterogeneously enhancing lesion measuring 28 x 24 x 30 mm in close relation to the posteroinferior aspect of the right lobe of thyroid gland with a few scattered non enhancing hypodense foci seen within the lesion. Tc99m sestamibi scan was obtained which showed intense abnormal focus of tracer concentration in the region of right thyroid lobe and showed persistence of the focus at four hours post injection CT image.

The familial associations of hyperparathyroidism were also ruled out. The patient was managed initially for biochemical correction of hypercalcaemia with adequate hydration, vitamin D supplementation and bisphosphonates (Zoledronic acid). After the stabilization of serum calcium and with preoperative localization of the hypersecretory right inferior parathyroid adenoma, the patient was taken up for a focused exploration and parathyroidectomy was performed. Postoperative recovery was uneventful. The parathormone dropped from preoperative levels of 532

pg/dL to 6 pg/dL in the post-operative evening (Table 2). The patient is still under the follow up at our hospital.

**Table 1: Highlighting important and out of range parameters (preoperative).**

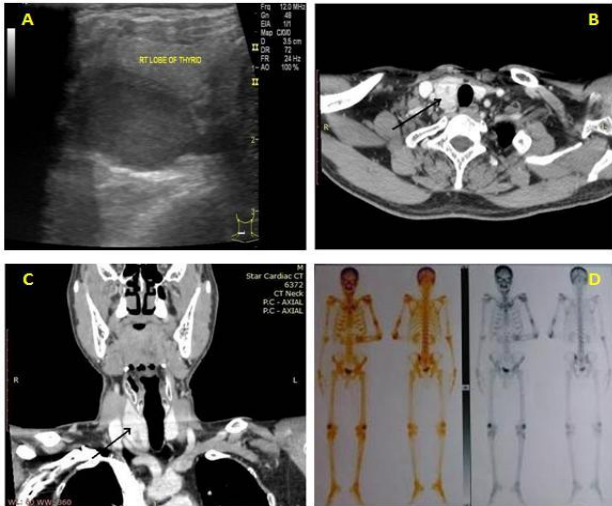
Parameter	Measured value
Haemoglobin	13.7 gm/dl
ALP	1077 IU/L
Serum calcium (Corrected)	12.6 mg/dL (8.8-10.5)
Serum phosphorus	4.3 mg/dL (2.5-4.5)
Parathormone	532 pg/dL (12-72)
Vitamin D3	18.9 ng/dL (30-100)
USG abdomen	Bilateral non obstructing renal calculi, largest 5.3 mm lower pole right kidney

**Table 2: Postoperative evening value of calcium, phosphorus and parathormone.**

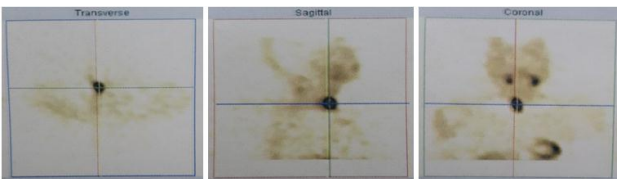
Parameter	Measured value
Parathormone	6 pg/dL (12-72)
Serum calcium (corrected)	9 mg/dL (8.8-10.5)
Serum phosphorus	4.6 mg/dL (2.5-4.5)



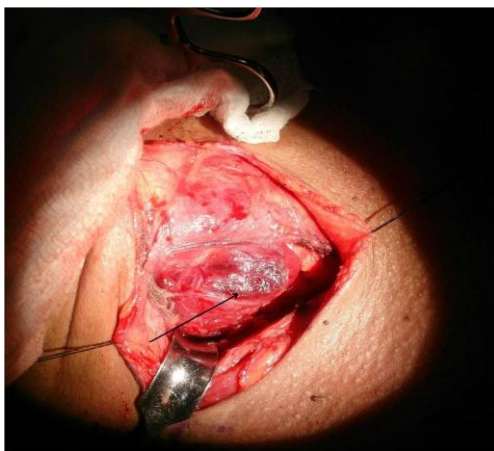
**Figure 1: (A) Gross osteopenia and Brown’s tumor with pathological fracture of left humerus, (B) Radiograph showing old fractured ulna with in-situ K-wire, (C) Osteitis fibrosa cystica: Characterized by fibrous degeneration of bone and formation of cystic cavities seen here at the ends of metacarpals, (D) Radiograph of pelvis showing generalized osteoporosis.**



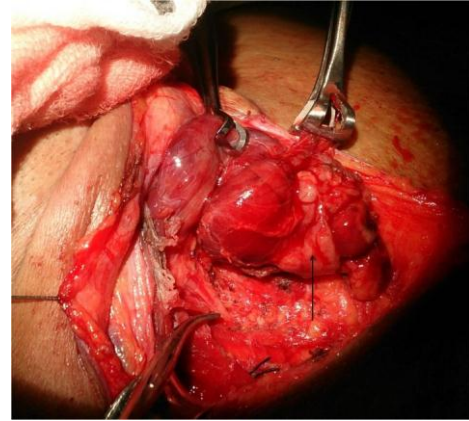
**Figure 2:** (A) USG neck revealing an hypo echoic lesion on the posterior aspect of the right lobe of the thyroid gland, (B) & (C) Contrast enhanced CT of the neck showing a heterogeneously enhancing lesion on the poster inferior aspect of right lobe of thyroid gland measuring 28 x 24 x 30 mm, (D) Whole body bone scan showing a patchy tracer uptake in entire skeleton with generalized osteoporosis.



**Figure 3:** Tc99m sestamibi scan SPECT images taken at 20 minutes, 1 hour, 2 hours and 4 hours showing intense abnormal focus of tracer concentration in the region of right lobe of thyroid gland. There was persistence of the intense abnormal focus of tracer till 4 hour image. The findings were suggestive of likely large inferior parathyroid adenoma.



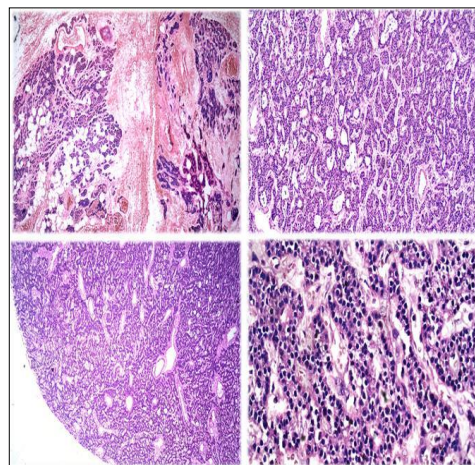
**Figure 4:** Focused approach for right inferior parathyroid gland adenoma. Exposure of the right lobe of the thyroid gland.



**Figure 5:** Intraoperative image showing the adenoma adherent to the thyroid lobe (arrow). Recurrent laryngeal nerve was identified and preserved (Artery pointing).



**Figure 6:** Resection specimen. The entire parathyroid adenoma was excised in toto. The specimen weighed 10 g.



**Figure 7:** Scattered group of tumor cells have markedly enlarged hyperchromatic nuclei with large nucleoli and abundant eosinophilic cytoplasm. Seen here in different magnification fields.

## DISCUSSION

Primary hyperparathyroidism is due to adenoma of the parathyroid gland in 85% of cases, the remainders are due to multiple adenomas and parathyroid cancers in that order of decreasing frequency. Secondary hyperparathyroidism is due to chronic vitamin D deficiency commonly due to chronic renal failure. Tertiary hyperparathyroidism is a result of autonomous parathyroid stimulation following persistent parathyroid stimulation. Primary hyperparathyroidism is commoner in females and the peak age incidence is between 30-50 years which increases with age. In USA, the annual incidence in >60 years is 0.2%.<sup>6</sup> Primary hyperparathyroidism usually presents with features of hypercalcaemia and the incidence of pathological fractures is relatively rare accounting for around 10% in two large series.<sup>7</sup> The radiographic features are seen in less than 2% of patients and include diffuse osteoporosis, subperiosteal erosions, Brown's tumors, pathological fractures and salt and pepper mottling of skull bones.<sup>6</sup> Extensive bone involvement and pathological fractures as presenting symptom has also been described.<sup>8</sup> Co-existent vitamin deficiency in patients with hyperparathyroidism may place them at significant higher risk of losing bone mineral deficiency. Two different types of bony involvement are described, the slowly progressive type leading to cortical thinning and osteoporosis and the other is rapidly progressive type.<sup>9</sup> The average weight of adenomas in patients who do not have significant bone disease is around 1 g with many less than 0.5 g, whereas tumors tend to be larger in patients with significant bone disease.

The parathyroid surgery has evolved over the years from traditional open all four gland exploration to minimally invasive focused or endoscopic parathyroidectomy.<sup>10</sup> The same has been made possible because of development of preoperative localization techniques which includes high resolution radiographic techniques and rapid intraoperative parathormone assay. However, USG and the Tc99m sestamibi scan are the most commonly employed methods for demonstrating parathyroid lesions. The sensitivity and specificity of detection of an adenoma with these approaches is almost similar as seen in comparative studies. Tc99m sestamibi is the commonest tracer used for imaging the parathyroid.<sup>11</sup> There is relative avid tracer uptake into the metabolically active adenomatous gland and the uptake persists in the hypersecretory gland on delayed images whereas the tracer washes out of rest of the less metabolically active parathyroids and the thyroid gland. It indicates the parathyroid gland(s) which is hyper-secreting and also its anatomical position which is variable.

The development of intraoperative parathormone assay (IOPTH) was an important advancement in surgical management of hyperparathyroidism as this could accurately refute the preoperative localization studies.<sup>12</sup> The success of this approach is based on the fact that

PTH has a half-life of 05 minutes. A drop in intraoperative parathormone levels of >50% from the baseline or pre-surgical excision value at 10 min after gland excision is accepted as the criterion by which the surgeon could conclude that all abnormal glands had been removed, without any need to identify all four glands. A six month follow up study from University of Miami has shown the success rate of 98% with the focused approach.<sup>13</sup> However, in some studies, a false positive drop of IOPTH has been documented in 2-3% of cases and also a similar proportion of cases have shown a false negative drop of PTH in 10 minutes.<sup>14,15</sup>

## CONCLUSION

This case illustrates the rare presentation of a large parathyroid adenoma in a young patient and the successful management of complications.

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