# Parathyroid Adenoma with Unusual Presentations of Rib Bone and Thoracic Vertebrae Fractures in a Premenopausal Female in Ibadan, Nigeria

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## Abstract

Parathyroid adenoma is the most common cause of primary hyperthyroidism which leads to abnormal calcium homeostasis, hypercalcemia, and reduction in bone density. A 37-year-old female referred from a private health facility with a 1-year history of upper back swelling and pain. The pain was worse when sitting down for long periods and with movement and relieved by rest. There was no antecedent history of trauma, but the patient had noticed poor appetite and weight loss. There were no constipation, no abdominal discomfort, and no symptom suggestive of hyperthyroidism or hypothyroidism. General physical examination revealed kyphoscoliosis, and vital signs were within normal limits. Spine X-ray showed features of cervical spondylosis. Computed tomography (CT) scan and magnetic resonance imaging showed pathologic fractures of the right 9<sup>th</sup> rib, anterior wedge compression, and reduction of T4 vertebrae with other abnormalities at T4–T5, T5–T6, T7–T8, T10–T11, and L4–L5 vertebrae. Bone marrow aspiration and serum electrophoresis were within normal limits. Serum calcium showed hypercalcemia. A CT scan of the neck was done which showed features of a right superior parathyroid adenoma. Blood count, other serum electrolytes, and thyroid function tests were all normal. A parathyroidectomy with right thyroid lobectomy was done. Histopathological examination of the resected parathyroid gland showed a diagnosis of parathyroid adenoma. A high index of suspicion is needed to diagnose this unusual presentation of parathyroid adenoma. Radiological imaging is an important tool for early diagnosis.

Keywords: Adenoma, atypical presentation, hyperparathyroidism, rib fracture

### INTRODUCTION

Primary hyperparathyroidism (PHPT) is a disorder caused by the excessive production of parathyroid hormone (PTH) leading to abnormal calcium homeostasis and hypercalcemia as well as an increase in osteoclastic activity with concomitant reduction in bone density.<sup>1</sup> The most common underlying cause of primary hyperthyroidism are parathyroid adenomas (80%–85%), followed by parathyroid hyperplasia, multiple adenomas, atypical adenomas, and very rarely parathyroid carcinoma.<sup>2</sup>

Clinical presentation is variable and nonspecific and may include weakness, nausea, constipation, severe bone, and kidney disease.<sup>3</sup> However, patients are commonly asymptomatic and diagnosed incidentally on imaging or as an incidental finding of raised serum calcium.<sup>3</sup> Osteitis fibrosa

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cystica is a rare complication characterized by osteoclastic bone resorption and formation of cyst-like brown tumors in and around the bone.<sup>4</sup>

Bone disease, if present, is rarely overt.<sup>5</sup> Radiographic manifestations are seen in <2% of all patients, and these include subperiosteal erosions, diffuse osteoporosis, cystic lesions, pathological fractures, "salt and pepper" mottling of

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the skull, and loss of lamina dura in the mandible.<sup>6</sup> Pathologic bone fractures, although uncommon, have been described in the literature sequel to PHPT, but they suggest both a late presentation and very severe disease.<sup>5</sup>

# **CASE REPORT**

A 37-year-old female referred from a private clinic to our hospital. She first noticed upper back swelling a year earlier, later developing upper back pain that radiated down the spine and lower limbs about 8 months later. The pain was worse when sitting down for long periods and with movement and relieved by rest. There was no antecedent history of trauma, but the patient had noticed poor appetite and weight loss. There were no constipation, no abdominal discomfort, and no symptom suggestive of hyperthyroidism or hypothyroidism. Examination revealed kyphoscoliosis, and her vital signs were normal.

Spine X-ray done showed features of cervical spondylosis. Computed tomography (CT) scan and magnetic resonance imaging (MRI) showed pathologic fractures of the right 9<sup>th</sup> rib, anterior wedge compression, and reduction of T4 vertebrae with other abnormalities at T4–T5, T5–T6, T7–T8, T10–T11, and L4–L5 vertebrae. Bone marrow aspiration and serum electrophoresis were normal. Further investigations revealed elevated serum calcium – 3.48 mmol/l (normal range is 2.15–2.65 mmol/l) and hyperparathyroidism (intact molecule – 90 pg/ml, normal range is 10–65 pg/ml). A CT scan of the neck was done which showed features of a right superior parathyroid adenoma [Figure 1]. Blood count, other serum electrolytes, and thyroid function tests were all normal.

She was admitted and worked up for a parathyroidectomy with right thyroid lobectomy. At surgery, the right superior parathyroid gland was enlarged, and this was resected. The other parathyroid glands were within normal limits. The thyroid gland also appeared normal.

The mass measured  $4.0 \text{ cm} \times 3.0 \text{ cm} \times 1.5 \text{ cm}$  and weighed 13 g. It was firm and multinodular, and the cut surfaces were homogenous grayish-yellow in appearance with multiple cystic spaces.



**Figure 1:** Computed tomography scan of the neck showing an enlarged right superior parathyroid gland

Histologic sections of the parathyroid gland mass showed a well-circumscribed benign neoplasm composed of monomorphic cells disposed in solid nests and follicular and pseudopapillary patterns. There was also a rim of compressed normal parathyroid gland tissue at the periphery. A diagnosis of parathyroid adenoma was made [Figure 2].

Immediate postoperative serum calcium was also normal (12.3 mmol/l). The patient subsequently had conservative management of the rib fracture and surgical management of the T4 thoracic spine wedge compression and was discharged home. The patient is doing well and on regular follow-up clinic visits.

## DISCUSSION

PHPT is a well-recognized clinical entity identified more than a century ago by Von Recklinghausen.<sup>7</sup> The disease results from excessive secretion of PTH either due to solitary (50%–85%) or multiple (10%) adenomas, hyperplasia (10%–40%), or rarely due to a carcinoma of a single parathyroid gland.<sup>8</sup> This condition is more common in females, and the incidence increases gradually with age, with a peak age of incidence ranging from the 4<sup>th</sup> to 6<sup>th</sup> decade.<sup>7</sup> In the USA, the annual incidence is around 0.2% in patients aged 60 years and above.<sup>5</sup> The age of the patient in this index case is 37 years. Extensive bony involvement with pathological fractures as a presenting feature of parathyroid adenoma has been documented, while some cases secondary to parathyroid carcinoma have also been described.<sup>8</sup> The patient in this index case had a solitary adenoma of the right superior parathyroid gland.

Two distinct types of bone lesions have been described in PHPT: the slowly progressive type which results in cortical thinning and osteoporosis and the rapidly progressive type.<sup>7</sup> Pathological fractures may occur in a weakened long bone or due to a cystic bone defect.<sup>7</sup> Co-existing Vitamin D deficiency has also been described in some patients.<sup>7</sup> The patient described in this index case had a slowly progressive type of bone lesion.

A principal test at present is the "immunoassay" for PTH I-84 as it distinguishes the hypercalcemia of malignancy from that of hyperparathyroidism.<sup>7</sup> However, this test was not done due to nonavailability. Clinically, hypercalcemia with serum PTH levels within or above the upper limits of normal and increased parathyroid cell mass suggests a diagnosis of primary hyperthyroidism.<sup>9</sup> PTH assays can also reliably distinguish PHPT from other causes of hypercalcemia.



**Figure 2:** (a-c) Photomicrographs showing parathyroid adenoma (H and E,  $\times$ 40 [a and b],  $\times$ 100 [c])

Once the diagnosis of PHPT has been made by biochemical analysis, the site or sites of adenomatous or hyperplastic parathyroid tissue must be identified.<sup>10</sup> CT scan, MRI, and thallium subtraction scans are useful for detecting parathyroid pathology in normal as well as ectopic locations.<sup>11</sup> In the index case, a CT scan of the neck was done which revealed a solitary parathyroid adenoma. Improvement in bone density, fracture healing, and prevention of further pathological fractures after successful parathyroid adenectomy has been demonstrated in this index case.

## CONCLUSION

A high index of suspicion is needed to diagnose this unusual presentation of parathyroid adenoma. A combination of biochemical tests (serum levels of calcium, phosphate, alkaline phosphatase, and parathormone assay) and radiological imaging will help in diagnosing PHPT. All patients with PHPT should also have Vitamin D level assessment in order to exclude the coexistence of Vitamin D deficiency with PHPT. Management of pathological fractures due to parathyroid adenoma includes surgical excision of the adenomatous parathyroid gland, calcium supplementation, and surgical management of the fractures.

#### **Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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#### **Conflicts of interest**

There are no conflicts of interest.

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