

# Concurrent parathyroid carcinoma and adenoma: A rare presentation of a rarer disease entity

Shamita Chatterjee, Udipta Ray, Shahana Gupta, Arghya Basu

Department of Surgery, Medical College, Kolkata, India

### ABSTRACT

Parathyroid carcinoma is a rare disease. But multiglandular parathyroid neoplasm is even rarer. A high level of suspicion, on the basis of clinical, hematological tests and intraoperative findings is necessary to treat this disease entity, particularly in the absence of palpable neck masses. Preoperative localization is important. Bilateral neck exploration should be done routinely and all 4 glands seen to avoid missing out other pathological glands.

**Key words:** Parathyroid adenoma, parathyroid carcinoma, multiglandular neoplasm, bilateral neck exploration

## INTRODUCTION

Parathyroid carcinoma is a rare disease. Usually thought to involve a single gland, it has been rarely seen to have multiglandular involvement. Though it commonly presents as a neck mass, the diagnosis presents a major challenge when it presents without any palpable neck mass. Though there has been a shift in the treatment paradigm to minimally invasive parathyroid surgery, bilateral neck exploration is advised in cases where parathyroid carcinoma is suspected to avoid missing out a second diseased gland. We report a rare case of suspected primary hyperparathyroidism, which turned out to be a case of multiglandular parathyroid neoplasm with carcinoma on one side and adenoma on the contralateral side.

## CASE REPORT

A 45-year-old male patient presented to the Medicine out patients' department with history of gradually progressive

generalized body pain, with severe pain in multiple joints of the body for past four months. He was advised to get investigated and given symptomatic treatment. The patient lost to follow up. He was brought to the emergency of the hospital a month later, with history of inability to move both lower limbs and the right upper limb for last five days.

The patient was admitted, and diagnosed to have bilateral femoral shaft fractures and left fracture shaft humerus. Radiological skeletal survey revealed gross osteoporosis, bilateral shaft femur fractures, fracture of the left humerus, multiple rib fractures on right side, tufting of the phalanges of the metacarpals of both hands and mottling of the skull bones [Figure 1].

Hematological examinations were within normal limits, except a hemoglobin level of 8.5 g/dl. Serum urea, creatinine, sodium and potassium were within normal limits. But serum calcium was elevated to a level of 14.9 mg/dl, while serum phosphate was 2.4 mg/dl. His 24 hours urinary calcium was 1176 (Normal – 100-300). Bone marrow biopsy was also within normal limits. Serum parathyroid hormone was 1842 pg/ml (Normal – 15-65 pg/ml). Primary hyperparathyroidism was suspected and an ultrasonography of the neck done. Though clinically the neck was normal, ultrasonography of the neck [Figure 2] revealed a pair of hypoechoic well defined SOLs abutting both lobes of thyroid from the posterior aspect. The one

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**Corresponding Author:** Dr. Shamita Chatterjee, 7 Bepin Pal Road, Kolkata - 700 026, India. E-mail: [dr\\_shamita@yahoo.com](mailto:dr_shamita@yahoo.com)

on the right side was  $3.6 \times 2.6$  cm, and the one on the left was  $1.5 \times 1.2$  cm in size. Both lobes of thyroid were normal and there was no evidence of lymphadenopathy. Technetium Tc99 m sestamibi scan revealed increased uptake consistent with the parathyroid gland only on the right side.

Bilateral neck exploration was planned. During exploration, a hard greyish mass of around  $4 \times 2$  cm was seen in the area of the right inferior parathyroid gland, densely adherent to the right lobe of thyroid. Tissue from the mass was sent for frozen section and reported to be parathyroid adenoma. But in view of clinical suspicion, intraoperative findings and hard grating sensation on cutting through the mass, an en block resection of the right thyroid lobe with the parathyroid mass and adjacent soft tissues was done. On the left side the left lower parathyroid gland was seen to be enlarged to  $1 \times 2$  cm approximately. It was firm and could be easily dissected free from the left lobe of thyroid. This gland was also excised. No lymphadenopathy was seen in the region. The upper two glands were identified and found to be normal. Intraoperative PTH assessment was not done as all four glands were identified. Post-operatively, the serum PTH came down to 16.2 pg/ml.

The post-operative period was uneventful and the serum calcium levels returned to normal levels within four days. The final histopathology revealed carcinoma of the right sided gland with capsular and vascular invasion. There was invasion into the adjacent thyroid lobe. The left lobe was reported to be a parathyroid adenoma [Figure 3].

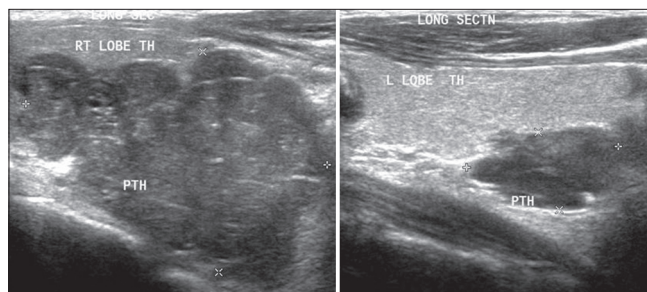
Anemia improved and was 10 g/dl a week after surgery. The fractures were treated conservatively initially in view of the gross osteoporosis. The patient was started on bisphosphonates. Definitive fixation of the fractures was done a month later, after the bone density improved.

## DISCUSSION

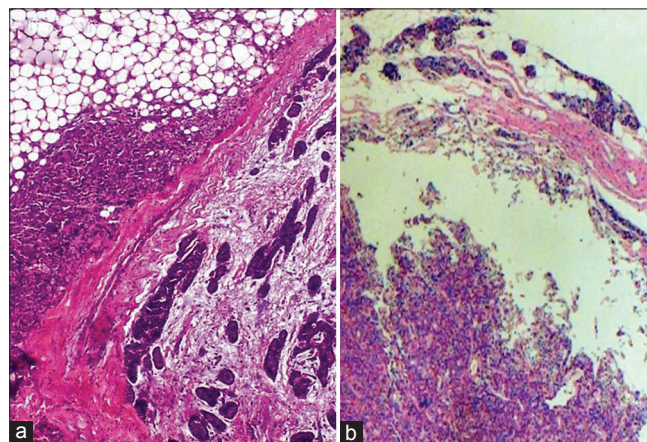
Parathyroid carcinoma is a rare endocrine malignancy and accounts for less than 1% of all cases of primary hyperparathyroidism.<sup>[1]</sup> The diagnosis becomes particularly challenging when no neck masses are palpable. It may be noted that a palpable neck mass is seen in only 30-76% of patients of parathyroid carcinoma, while fewer than 5% of patients with adenomas have a palpable mass in the neck. The diagnosis is based on a strong clinical suspicion, supported with high levels of serum calcium ( $>14$  mg/dl) and highly elevated levels of serum parathyroid hormones ( $>5$  times the normal levels).<sup>[2]</sup>



**Figure 1:** Radiographs of left femur showing pathological fracture, right humerus with gross osteoporosis, left humerus with pathological fracture



**Figure 2:** Ultrasonography of the neck showing bilateral hypoechoic SOLs of the parathyroid glands



**Figure 3:** Hematoxylin eosin stained high-power microscopic view of (a) the right parathyroid gland showing parathyroid carcinoma with evidence of vascular invasion and invasion into surrounding thyroid tissue. (b) the left parathyroid gland showing evidence of parathyroid adenoma

Imaging is mandatory for preoperative localization of the glands. Ultrasonography and Tc99m sestamibi scans are more commonly done, though the sensitivity of ultrasound can be as low as 65%.<sup>[3]</sup> In multiglandular disease, the detection rate is often lower. In our case, though both glands were picked up by ultrasonography, the left one was not detected by Tc99m sestamibi scan.

Intraoperative suspicion is very important to make a diagnosis of carcinoma. Size, color, consistency, adherence and infiltration into surrounding organs often lead to the suspicion of carcinoma. Though frozen section is useful,<sup>[1,4]</sup> it is often negative, as in our case, and should not bias our judgment against malignancy.

Surgery is the treatment of choice. The involved parathyroid gland with ipsilateral lobe of thyroid and any soft tissue to which the gland was adherent should be excised en block. Whether prophylactic ipsilateral neck dissection is warranted is debatable.<sup>[5]</sup> In our case, we did not do a neck dissection as clinically and sonologically no neck nodes were found.

Anemia in primary hyperparathyroidism may be due to various causes, but bone marrow fibrosis is a very significant cause. The anemia and marrow fibrosis in these patients improve following parathyroidectomy.<sup>[6]</sup> Though our patient did not show evidence of significant marrow fibrosis on bone marrow biopsy, his anemia improved following surgery.

Parathyroid carcinoma may be associated with familial hyperparathyroidism, in patients suffering from MEN I, in patients of hyperparathyroidism jaw tumor syndrome or in patients with chronic renal failure. Our patient did not have any history or features suggestive of the above. Hence clinical and biochemical suspicion were of paramount importance.

Parathyroid carcinoma may be associated with hyperplasia of other parathyroid glands.<sup>[7]</sup> But it is rarely associated with adenoma of the other parathyroid glands or carcinoma of another ipsilateral or contralateral parathyroid gland.<sup>[8-10]</sup> Our patient had carcinoma and adenoma on either side. It is mandatory to do a bilateral neck exploration, even if imaging of the other side is apparently normal, to avoid missing out on these diseased glands.

## CONCLUSION

Multiglandular parathyroid neoplasm is a very rare entity. A high level of suspicion, on the basis of clinical, hematological tests and intraoperative findings is necessary to treat this disease entity. In the era of minimally invasive surgeries, if parathyroid carcinoma is suspected, bilateral neck exploration should be done routinely and all four glands seen to avoid missing out other pathological glands.

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