

# Ectopic Parathyroid Adenoma in the Carotid Sheath

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## Abstract

Parathyroid adenomas can be found in various ectopic positions including the carotid sheath. We report a 52-year-old Libyan woman who presented with neck mass and a fragility fracture. Her serum Calcium was elevated (14.3 mg/dl), as well as her parathyroid hormone (1202 pg/ml). Technetium-99m-sestamibi scan showed a large localized parathyroid adenoma at the upper lateral left aspect of the neck just below the left submandibular gland. The patient underwent surgical resection, and the histopathology confirmed the diagnosis of parathyroid adenoma. The rarity of the ectopic parathyroid adenoma in the carotid sheath led to a delayed diagnosis of this case. Therefore, ectopic parathyroid adenoma should be considered in the differential diagnosis of masses that arise from the carotid sheath.

**Keywords:** Adenoma, carotid body tumor, carotid sheath, ectopic, hyperparathyroidism, parathyroid, sestamibi

## INTRODUCTION

Parathyroid glands are four small-sized glands, located behind the upper and lower poles of the thyroid gland. The two superior glands are usually located at the posterolateral aspect of the upper pole of the thyroid whereas the inferior glands are found frequently near the lower thyroid pole. However, parathyroid glands can be found in ectopic positions in 6%–16% of normal adults.<sup>[1]</sup>

These ectopic positions include intrathyroidal,<sup>[1,2]</sup> carotid sheath,<sup>[1]</sup> retroesophageal sites, mediastinal sites, and thymus.<sup>[1-3]</sup> Compared to the superior parathyroid glands the inferior glands demonstrate a greater degree of variability in location since they might stopover anywhere along the thyrothymic tract as they descend with the thymus gland from the angle of the mandible down to the pericardium during embryogenesis.<sup>[4]</sup>

## CASE REPORT

A 52-year-old Libyan woman was admitted to the orthopedic department with a fragility fracture of her left hip following minor trauma. An endocrine consultation was requested. On further inquiring, she gave a 1-year history of painless left neck swelling that is gradually increased in size and was diagnosed as carotid body tumor based on ultrasonic and computed tomography (CT) scan appearance. She also described an increasing thirst and urination over the last

year. Two months back, she started complaining of muscle pain and cramps in both lower limbs. Her past medical history and family history were unremarkable. Her neck examination shows painless, nonpulsatile left neck swelling at the carotid artery moving in a vertical plane but not the horizontal plane. Initial laboratory data revealed: serum calcium: 14.3 mg/dl, serum phosphorous: 1.84 mg/dl (2.7–4.5), parathyroid hormone (PTH): 1202 pg/ml (15–65), alkaline phosphatase: 325 u/L (30–120), and vitamin D3: 12.89 ng/ml. Neck ultrasound displayed a well-demarcated hypoechoic lesion at the bifurcation of the left carotid artery measuring 3 cm × 2.5 cm. Magnetic resonance imaging of the neck showed 2.5 cm × 3.5 cm × 2 cm oval-shaped well-defined mass lesion on the left submandibular region abutting the lateral aspect of the left carotid sheath just at the level of the carotid bifurcation. The lesion displayed T1 hypointensity and T2 hyperintensity signal together with homogenous enhancement following the intravenous (IV) gadolinium administration. There was no obvious parathyroid adenoma at the normal locations. Technetium-99m-sestamibi scan of the parathyroid multiplexed ion beam imaging (MIBI) showed a large localized

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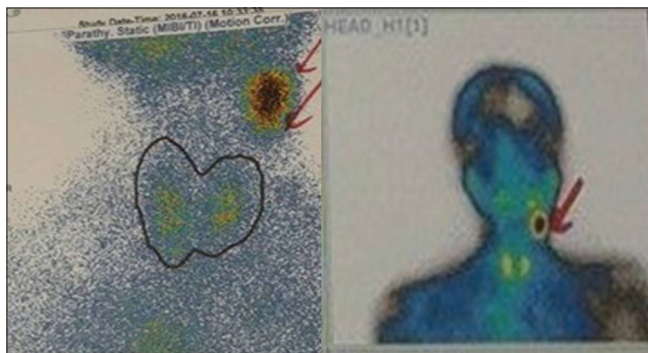
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parathyroid adenoma at the upper lateral aspect of the left neck just below the left submandibular gland [Figure 1]. CT scan of the pelvis shows multiple cystic lesions of the hip bones consistent with osteitis fibrosa cystica [Figure 2].

A provisional diagnosis of primary hyperparathyroidism with advanced bone disease due to possibly an ectopic parathyroid neoplasm was made. Due to the unavailability of surgical intervention at the time of diagnosis, she was maintained on cinacalcet 30 mg BD, risedronate once/week. Three weeks later, the calcium dropped to 10.4 mg/dl, and PTH dropped to 917 pg/ml. Later on, she underwent neck exploration with resection of the above-mentioned mass, after which her PTH fell to 1.6 pmol/L (1.6–6.9), and she developed a hungry bone syndrome which required treatment with IV calcium infusion for few days followed by oral calcium carbonate and alfacalcidol which reduced gradually according to her laboratory data to be stopped completely 9 months later. The histopathology confirmed the diagnosis of parathyroid adenoma weighting 8.9 g, with no evidence of malignancy.

## DISCUSSION

The most common cause of primary hyperparathyroidism is PTH secreting adenoma, which represents around 80% of the cases. Due to the embryological migration of the parathyroid gland, it can be found anywhere along the thyrothymic tract, most commonly with the thymus<sup>[5]</sup> and rarely within the carotid sheath as it is described in this case. Roy *et al.* reported the most extensive series of ectopic parathyroid adenomas with 202 patients, out of those the most common site for the ectopic glands was within the thymus gland (38%), followed by a retro/paraesophageal region (31%), intrathyroid sites (18%), mediastinal sites (6%), and the carotid sheath (3%).<sup>[3]</sup> Another two smaller studies reported 37<sup>[5]</sup> and 102<sup>[6]</sup> patients, respectively, and the prevalence of ectopic adenoma in the carotid sheath in these reports was 7%<sup>[5]</sup> and 9%<sup>[6]</sup> respectively. Despite this variation in the frequency of ectopic parathyroid adenoma within the carotid sheath (3%–9%), all three studies indicate that the least common location is the carotid sheath.<sup>[3,5,6]</sup>



**Figure 1:** Technetium-99m-sestamibi scan of the parathyroid showing a large localized parathyroid adenoma at the upper lateral aspect of the left neck just below the left submandibular gland

Ectopic parathyroid adenoma can represent a diagnostic dilemma, in a scenario like our reported case, a case reported by Malm *et al.* described a mass which was suspicious for carotid body tumor and turned to be an ectopic parathyroid adenoma in the carotid sheath.<sup>[7]</sup> In an analysis of 202 patients with ectopic parathyroid adenoma, Roy *et al.* reported an overall sensitivity of preoperative MIBI scans and ultrasound scan for detecting ectopic glands of 89% and 59% respectively.<sup>[3]</sup> However, in the same analysis, there was no imaging modality with a high rate of detecting ectopic glands in the carotid sheath, as the ultrasound sensitivity was 0% and the MIBI scan sensitivity was only 20%.<sup>[3]</sup> The adenoma in our reported patient was detected readily by the MIBI scan, perhaps due to the large size of the adenoma in this patient.

Lack of a preoperative definitive diagnostic procedure makes the diagnosis of the ectopic parathyroid adenoma challenging, and it is a prevalent cause for persistence or recurrence of hyperparathyroidism.<sup>[8]</sup> The higher rate of success of the first surgical intervention is directly linked to the accurate localization of the gland using preoperative imaging, it is recommended to use one or more of the imaging studies for precise localization of the ectopic gland such as 99 m Tc-sestamibi scan, ultrasound, and four-dimensional-CT-scan.<sup>[9,10]</sup>

## CONCLUSIONS

The rarity of the ectopic parathyroid adenoma in the carotid sheath led to a delayed diagnosis of this case since the neck mass was thought to be a carotid body tumor. Ectopic parathyroid adenoma should be considered in the differential diagnosis of masses that arise from the carotid sheath.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient consented for her images and other clinical information to be reported in the journal. The patient understands that name and initial will not



**Figure 2:** Computed tomography scan of the pelvis showing multiple cystic lesions of the hip bones consistent with osteitis fibrosa cystica

be published, and outstanding efforts will be made to conceal identity, but anonymity cannot be guaranteed.

### Author's contributions

All authors contributed to the care of the patient, drafting of the case report, revision, and approval of its final version.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

### Compliance with ethical principles

No prior ethical approval is usually required for single-case reports at our institution

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