

A Case of Submandibular Ectopic Parathyroid Adenoma: Diagnostic Challenges and Clinical Insights

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We report a case of a left submandibular ectopic parathyroid adenoma (EPA) with a tortuous diagnostic course, highlighting potential misdiagnoses and key diagnostic strategies. A 51-year-old male office worker presented with an incidentally discovered left submandibular mass persisting for 6 months and a newly detected thyroid nodule for 1 month. Initially, the submandibular mass was misdiagnosed as reactive lymphadenopathy at a local clinic, where only physical examination was performed without imaging or serological tests. Five months later, another hospital misdiagnosed the same mass as a schwannoma based on neck ultrasound (Figure 1a), during which a TI-RADS 4A nodule in the upper pole of the right thyroid was incidentally identified. Contrast-enhanced computed tomography (CT) of the neck (Figure 1b) prompted referral for surgical evaluation.

The patient was referred to our thyroid surgery clinic due to the right upper thyroid TI-RADS 4A nodule, seeking simultaneous resection of both lesions. Comprehensive evaluation revealed elevated serum calcium (2.79 mmol/L; reference range, 2.10–2.55 mmol/L), decreased serum phosphorus (0.70 mmol/L; reference range, 0.81–1.45 mmol/L), and markedly elevated intact parathyroid hormone (iPTH, 547 pg/mL; reference range, 15–65 pg/mL), suggestive of primary hyperparathyroidism (PHPT). ⁹⁹PTH, methoxyisobutylisonitrile (MIBI) scintigraphy showed no abnormal uptake (Figures 1c, d). Ultrasound-guided fine-needle aspiration biopsy (FNAB) revealed epithelioid cells arranged in nested and sheet-like patterns, consistent with parathyroid adenoma, confirming left submandibular EPA. Genetic testing for *MEN1*, *CDC73*, and *AIP* genes was negative. Because FNAB for parathyroid lesions is rarely performed in our practice, we did not measure needle washout PTH, relying solely on cytological examination.

The patient underwent resection of the left submandibular EPA combined with right thyroid nodule excision. A 6-cm transverse incision was made along the suprasternal skin crease to facilitate exploration of remaining parathyroid glands and thyroid nodule removal. After incising the skin and subcutaneous tissue, the left sternocleidomastoid muscle was retracted laterally to expose the left

internal jugular vein and common carotid artery. Dissection along the common carotid artery revealed a mass measuring approximately 2 cm in diameter inferior to the left carotid bifurcation, adjacent to the left submandibular gland (Figures 1e, f). The mass was carefully mobilized to avoid injury to the vagus nerve, carotid artery, and internal jugular vein, while preserving the vascular pedicle of the upper thyroid pole. The mass was completely resected without iatrogenic injury. Three normal parathyroid glands (right superior, right inferior, and left inferior) were identified; no left superior parathyroid tissue was found in its normal anatomical location, indicating that the ectopic mass originated from the left superior parathyroid gland. The resected specimen appeared grayish-red and well-circumscribed (Figures 1g, h). Intraoperative frozen section suggested “parathyroid hyperplastic tissue, adenoma not excluded”; the right thyroid nodule was confirmed as a benign nodular goiter. Postoperatively, serum calcium and iPTH rapidly normalized (2.33 mmol/L and 13 pg/mL, respectively) without hypocalcemic symptoms. Final histopathology confirmed EPA (Figure 1i) and a benign thyroid nodule. Two-year follow-up showed no recurrence.

PHPT is a metabolic disorder caused by excessive PTH secretion. EPA accounts for 10–22%¹ of PHPT cases, commonly occurring in intrathyroidal and mediastinum;^{2,3} mediastinal sites; submandibular ectopy is extremely rare (4–6% of EPA cases,^{1,2} rendering), making preoperative diagnosis challenging. In this case, the sequential misdiagnoses were due to limited awareness of this rare ectopic site and over-reliance on empirical judgment, compounded by the patient’s asymptomatic presentation (without typical PHPT manifestations such as bone pain).⁴ MIBI scintigraphy, the first-line modality for localizing hyperfunctioning parathyroid tissue, may yield false-negative results in rare ectopic locations due to low tracer uptake or reduced lesion activity.⁵ For MIBI-negative cases, ¹⁸F-fluorocholine positron emission tomography (PET)-CT is a valuable adjunct, offering a reported detection sensitivity of 93.7% and superior target-to-background ratio.⁵ Although FNAB combined with serological tests confirmed the diagnosis in this patient, PET-



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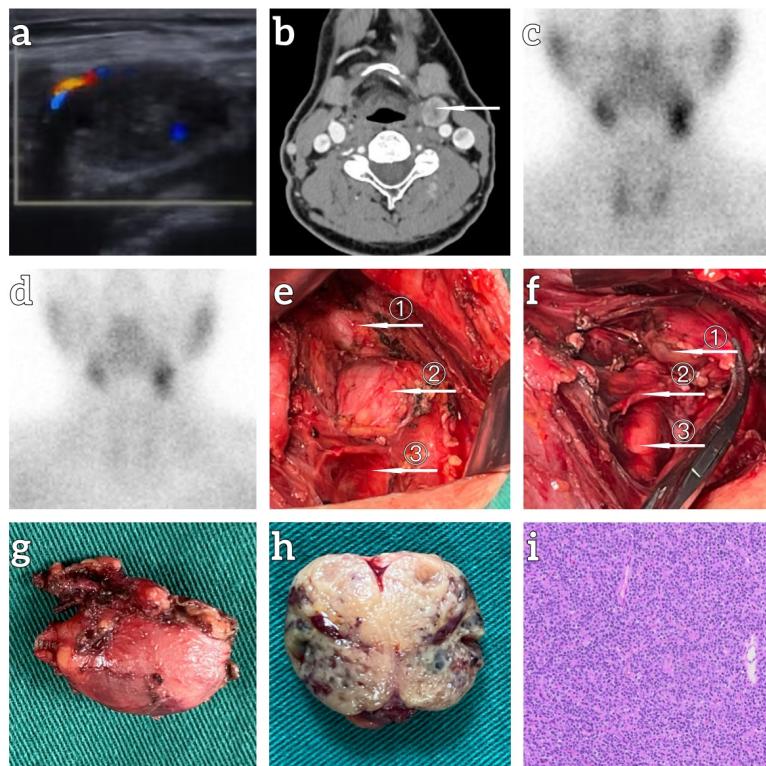


FIG. 1. Radiological, intraoperative, and histopathological findings of the case. Color Doppler ultrasound image showing a hypoechoic nodule adjacent to the left submandibular gland and medial to the left common carotid artery (a). Contrast-enhanced neck CT image: the arrow indicates the mass with moderately heterogeneous enhancement (b). ^{99}Tcm -MIBI scintigraphy images (c, d): Early phase (30 minutes post-injection) (c); delayed phase (2 hours post-injection) (d); no abnormal radioactive tracer accumulation was detected in the neck or mediastinum. Intraoperative exposure of the ectopic parathyroid adenoma: arrow ① = submandibular gland; arrow ② = ectopic parathyroid adenoma; arrow ③ = left common carotid artery (e). Intraoperative mobilization of the adenoma: arrow ① = ectopic parathyroid adenoma; arrow ② = superior thyroid vein; arrow ③ = left common carotid artery (f). Gross appearance of the resected adenoma specimen (g). Cut surface of the adenoma showing grayish-red solid tissue (h). Postoperative histopathological image (hematoxylin and eosin staining) (i). CT, computed tomography; MIBI, methoxyisobutylisonitrile.

CT can provide precise anatomical localization and metabolic information for lesions with occult locations or indeterminate FNAB results, guiding surgical planning.

For neck masses in special locations (e.g., carotid sheath, submandibular region), FNAB effectively differentiates parathyroid lesions from neurogenic tumors, lymph node disorders, and salivary gland neoplasms, significantly improving diagnostic accuracy.⁶ When ectopic parathyroid tissue is suspected, PTH measurement in needle washout fluid¹ can further confirm ectopic parathyroid tissue; in this case, cytology alone was sufficient due to our insufficient awareness of this approach. Delayed diagnosis of EPA may lead to complications such as osteoporosis, pathological fractures, and nephrocalcinosis, complicating surgical⁴ management. Therefore, comprehensive evaluation—including serology, FNAB, and appropriate imaging—is crucial for diagnosing neck masses in rare locations.

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Informed Consent: Written informed consent was obtained from the patient for the publication of this case report and any accompanying images. The patient has been informed that their personal identifying information will be anonymized to protect privacy.