

Odd Sites of Parathyroid Adenomas: 18F-fluorocholine PET/CT Pointing to the Right Place

Abstract

Primary hyperparathyroidism is mostly caused by parathyroid adenoma(s) which are generally localized using routine imaging modalities such as neck ultrasonography and ^{99m}Tc-SestaMIBI scintigraphy. However, these can miss ectopic parathyroid adenomas due to their limited sensitivities. These ectopic lesions can later lead to failure of surgical excision and necessitate the need for a re-exploration. ¹⁸F-fluorocholine (FCH) positron emission tomography/computed tomography (PET/CT) can help in the localization of these ectopic adenomas due to its superior detection rates and spatial resolution. Herein, we report a case of ectopic parathyroid adenomas that were localized on FCH PET/CT.

Keywords: ¹⁸F-fluorocholine positron emission tomography/computed tomography, ^{99m}Tc-SestaMIBI, ectopic parathyroid adenomas, neck ultrasonography

Introduction

Parathyroid adenomas are the most frequent cause of primary hyperparathyroidism (pHPT). Ectopic adenomas are found in 5%–10% of the cases.^[1] Surgical excision is the definitive management of pHPT but due to the unusual location of ectopic parathyroid adenomas and the limited operative field, bilateral neck exploration is invariably unsuccessful. This poses a challenge for preoperative localization.^[2] Here, we are reporting a case of ectopic parathyroid adenoma which is located in the submandibular region and was localized accurately on ¹⁸F-fluorocholine (FCH) positron emission tomography/computed tomography (PET/CT) following which the patient underwent successful surgical excision.

Case Report

A 32-year-old female is presented with generalized weakness, tiredness, backache, gastritis, and palpitations for a year. She underwent routine biochemical workup that confirmed hyperparathyroidism with an iPTH of 3039 pg/dL and serum calcium of 10 mg/dL with a serum creatinine of 3.2 mg/dL. DEXA scan was suggestive of osteoporosis. Localization attempts

with neck ultrasound ultrasonography (USG) and ^{99m}Tc-SestaMIBI scintigraphy with single-photon emission computed tomography/CT (SPECT/CT) were unsuccessful, following which an FCH PET/CT was done which localized a tracer-avid soft-tissue density lesion measuring 7 mm × 7 mm × 20 mm, starting posterior to the left submandibular gland, and extending inferiorly lateral to the left pyriform fossa up to the level of the left lamina of thyroid cartilage, medial to left sternocleidomastoid muscle which was consistent with a submandibular ectopic parathyroid adenoma [Figure 1]. A 3 and 1/2 gland parathyroidectomy with submandibular parathyroid adenoma excision was performed and histopathologically confirmed. Postoperatively, she developed hungry bone syndrome that was managed with intravenous calcium. Postdischarge, her serum calcium had normalized, and she was asymptomatic.

Discussion

Parathyroid glands are tiny endocrine glands (usually 4 in number) located in the vicinity of the thyroid gland. They arise from the embryonic pharynx alongside the thyroid and thymus. The inferior parathyroid glands and thymus arise from the third branchial pouch while

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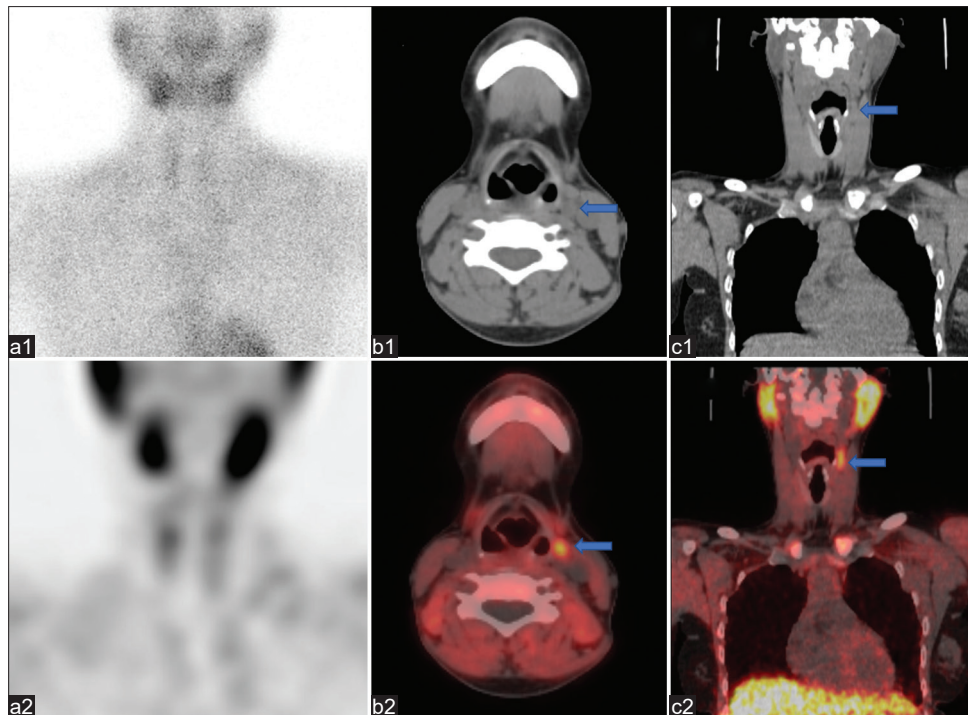


Figure 1: ^{99m}Tc-SestaMIBI scintigraphy planar (a1) and SPECT (a2) images reveal a negative scan and ¹⁸F-fluorocholine PET/CT axial (b1 and 2) and coronal (c1 and 2) images reveal a soft-tissue density lesion (blue arrow marked), starting posterior to the left submandibular gland and extending from lateral to the left pyriform fossa up to level of left lamina of thyroid cartilage just medial to left sternocleidomastoid muscle with increased tracer uptake. PET/CT: Positron emission tomography/computed tomography, SPECT: Single-photon emission computed tomography

the superior parathyroid glands arise from the fourth pharyngeal pouch along with the lateral thyroid gland. The inferior parathyroids descend downward almost up to the lower pole of the thyroid and have a greater migration as compared to the superior parathyroid glands. Due to their long course during development, they are commonly seen in ectopic locations. Some individuals also demonstrate supernumerary glands in about 2.5%–22%.^[3] The inferior parathyroid glands can have a wide range of locations, the most common being the posterolateral aspect of the inferior pole of the thyroid gland. Inferior glands can be found close to the thyrothymic ligament, anterior mediastinum, at the carotid bifurcation, be intrathyroidal or intrathyroidic.

Here, we report a case of ectopic parathyroid adenomas with difficult preoperative localization. Many a time, such patients undergo surgical interventions to no avail.

In the surgical treatment of hyperparathyroidism, precise preoperative localization of hyperfunctioning parathyroid tissue, including detection of ectopic parathyroid lesions, has become increasingly important, due to a paradigm shift from bilateral neck exploration to minimally invasive parathyroidectomy in the past few decades.^[4,5] Ultrasound neck and ^{99m}Tc-SestaMIBI scintigraphy, often in combination with SPECT/CT, are the first-line investigations. Various studies have reported the sensitivity of this approach to be between 77% and 89%.^[6] The sensitivity of both these modalities may be decreased in patients with multiglandular disease, multinodular

goiter, or ectopic parathyroid adenoma(s).^[5] Negative or inconclusive preoperative results increase the chances of surgical failure.

Therefore, to improve the detection of these pathological glands, it is necessary to add another imaging modality for proper localization.

FCH PET/CT is based on increased cell membrane turnover in imaging with parathyroid adenoma or hyperplasia which leads to increased choline uptake and phosphorylation by choline kinase into phosphatidylcholine. Hence, upregulation of choline kinase activity leads to increased fluorocholine uptake^[7,8] which was found to be more intense in adenomas compared to hyperplastic glands.^[9] FCH PET/CT provides higher spatial resolution, shorter scanning time, lesser radiation dose and has provided promising results with detection rates exceeding 90%.^[4] This investigation may not just be useful in patients with negative ^{99m}Tc-SestaMIBI scintigraphy but may even show additional lesions in patients with already positive ^{99m}Tc-SestaMIBI scintigraphy scan.^[7] Unlike ^{99m}Tc-SestaMIBI, FCH PET/CT can successfully localize small, hyperplastic, and multiple hyperfunctioning parathyroid glands, irrespective of their histopathological composition.^[10] FCH PET/CT has the potential to be a standard investigation in the detection of parathyroid lesions. Therefore, FCH PET/CT may play a key role as a problem-solving tool in difficult cases such as those with recurrent hyperparathyroidism and

ectopic lesions.^[10] In a prospective study of 105 patients with negative or discordant USG and MIBI results, the sensitivity, positive predictive value, and accuracy of FCH PET/CT in the detection of pHPT were 94.1%, 97.9%, and 92.4%, respectively.^[10] A recent meta-analysis evaluating the diagnostic performance of FCH PET/CT in hyperparathyroidism revealed a sensitivity and specificity of 90% and 94%, respectively.^[11] When FCH PET/CT is used as a first-line imaging modality, per patient and per lesion-based detection rates were 96% and 90%.^[4] Ectopic parathyroid adenoma in submandibular location is a rare entity and is only discussed in a few case reports.^[2,12]

In our case study with ectopic parathyroid adenoma, we found that FCH PET/CT was able to accurately localize the ectopic adenoma in this cases. The lesion was differentiated from lymph nodes by cross-confirmation with ultrasound. False negatives on ^{99m}Tc-SestaMIBI scans could be attributed to masking of MIBI uptake in the lesion due to intense physiological MIBI uptake in the submandibular gland in our case.

The high uptake of the FCH tracer with superior image contrast facilitated both reporting physicians to identify these lesions more convincingly as parathyroid adenoma.

Conclusion

Submandibular parathyroid adenomas are rare. The usual gold-standard operative management of bilateral neck exploration for parathyroid adenoma does not include these ectopic sites, and hence, it becomes essential to properly localize the parathyroid adenoma(s) preoperatively. In case of previous failed surgeries, FCH PET/CT plays an important role in adequate localization of culprit lesions in patients being considered for re-exploration.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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