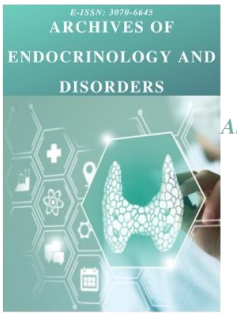

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Case Report

Retropharyngeal Ectopic Parathyroid Adenoma: A Case Resolved with 18F-Fluorocholine Positron Emission Tomography (PET)

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Abstract

Primary hyperparathyroidism is caused by either adenoma or hyperplasia involving the chief cells of parathyroid glands. Primary hyperparathyroidism is third most common endocrine disease affecting women 2-3 times more often than men. The incidence of Primary Hyperparathyroidism (PHPT) is increasing with rate of 42:100,000 per year. Both parathyroid adenoma or hyperplasia is surgically curable, but the real twist lies in pre-operative localization. The usual pre-operative localization of PHPT is done by two modalities namely cervical ultrasound and SestaMIBI scan. Cervical ultrasound is cheap and easily available and can simultaneously detect any thyroid pathology if present. The radionuclide imaging is particularly useful in identification of ectopic glands as well as easier recognition of posteriorly located upper glands. When both modalities are used together it increases the sensitivity of pre-operative localization of the glands.

Primary hyperparathyroidism due to ectopic parathyroid adenoma is seen in 16-23% of cases. We report a case of a 55-year-old female who presented with elevated calcium levels on routine investigations. She had bilateral renal calculi with a serum calcium level of 11.8 mg/dL and an iPTH level of 98.1 pg/mL. Ultrasound of the neck revealed a hyperechoic nodule in the inferior pole of the left lobe of the thyroid, suspicious for a parathyroid adenoma and sestamibi confirmed the lesion on right side. She underwent a right inferior parathyroidectomy, but her iPTH levels were persistently elevated postoperatively. She was further evaluated for hyperparathyroidism with an 18-F fluorocholine Positron Emission Tomography (PET) scan, which revealed a right retropharyngeal parathyroid adenoma. She underwent re-exploration of the neck and removal of the parathyroid adenoma. Intra-op iPTH dropped to normal levels. The histopathological findings were consistent with parathyroid adenoma. Patient is in follow-up with normal calcium and iPTH levels.

Keywords: Primary Hyperparathyroidism; Ectopic Parathyroid Adenoma; 18F-Choline PET Scan; iPTH; Nuclear Imaging

Introduction

Primary hyperparathyroidism is caused by either adenoma or hyperplasia involving the chief cells of parathyroid glands. Primary hyperparathyroidism is third most common endocrine disease affecting women 2-3 times more often than men [1]. The incidence of Primary Hyperparathyroidism (PHPT) is increasing with rate of 42:100,000 per year [2]. Primary Hyperparathyroidism (PHPT) is commonly seen in women in 5th decade and can be associated with radiation exposure,

sarcoidosis, hereditary and genetic syndrome hyperparathyroidism-jaw tumor syndrome, multiple endocrine neoplasia or isolated familial hyperparathyroidism [3]. Approximately 70-80% of the patients with benign PHPT are asymptomatic and detected incidentally due to elevated Calcium levels. The remaining patients present with classical symptoms and sign of PHPT. The most common site for ectopic parathyroid gland is thymus and superior mediastinum. the incidence of ectopic parathyroid gland is 23.8%. Ectopic left lower parathyroid gland is 13.2%. Left lower parathyroid gland is most prone for ectopia, mostly located in thymus or superior mediastinum [4]. Pre-operative use of Tc99m MIBI SPECT/CT and ultrasound is found to be sensitive to detect PHPT in 92.31% which is higher than either investigation modality.

The recurrence of hypercalcemia post-operatively is higher than previously thought. Most cases are transient and often associated with other factors. The late recurrence is found to be 15%. Because of this long term follow-up should be considered [5]. The persistent PHPT is defined as failure to achieve normocalcemia and normal intact Parathyroid Hormone (iPTH) levels within 6 months of parathyroidectomy [1]. In our case vigilant follow-up leads to early detection of persistent post-operative hypercalcemia and choline PET imaging helped us to localize the exact site of the lesion, thus enabling us for improved patient care.

Case Report

A 55 years old female, known case of Hypertension for 10 years, was on Cilnidipine+ Metoprolol (40/50)- 1 tab once daily. Presented to our outpatient center with accidentally detected elevated calcium levels. Patient complained of generalized weakness. There was no history of bony growths or bone fractures. Examination revealed weight 59.8 kg, height-157 cm with BMI- 24.26 kg/m^2. Blood pressure 145/93 mmHg, Pulse rate -90/min. No osteitis fibrosa cystica noted. No anterior neck swelling noted. Systemic examination was found to be normal. Initial evaluation revealed Calcium -11.8 mg/dl, iPTH- 98.1 pg/ml; TSH- 2.13 uIU/ml. Ultrasound abdomen revealed bilateral renal calculi. Ultrasound neck revealed complex cystic, hyperechoic, 14×12 mm, nodule in inferior pole of left lobe of thyroid. Parathyroid scintigraphy revealed-persistent faint tracer activity noted in inferior pole of right lobe of thyroid, 0.2×0.3 cm, suggestive of right inferior parathyroid adenoma. Patient was subjected to neck exploration and right inferior parathyroidectomy. Intra-op iPTH fall to 73 pg/ml. The frozen section showed parathyroid adenoma. Subsequently in follow-up Ca 9.9 mg/dl; iPTH 86.6 pg/ml and at 8 weeks Ca 10.4 mg/dl and iPTH 92.2 pg/ml. This persistently rising trend in iPTH with 103 pg/ml; Ca 10.8 mg/dl. lead to repeat imaging with F18- Fluro choline PET. This FCH PET revealed right retropharyngeal parathyroid adenoma of size 10×6 mm with vertical extension of 19 mm. With this new finding and persistently elevated iPTH levels, patient was subjected to re- exploration of neck and removal of parathyroid adenoma. Intra-op iPTH dropped to 12.5 pg/ml. Patient did not have any intra-operative or immediate post-operative complications following re-exploration surgery (Fig. 1-6).

Follow-up

Histopathology finding of surgical specimen corresponds to parathyroid adenoma. Patient is in follow-up with normal calcium (8.8 mg/dl) and iPTH levels (20 pg/ml).

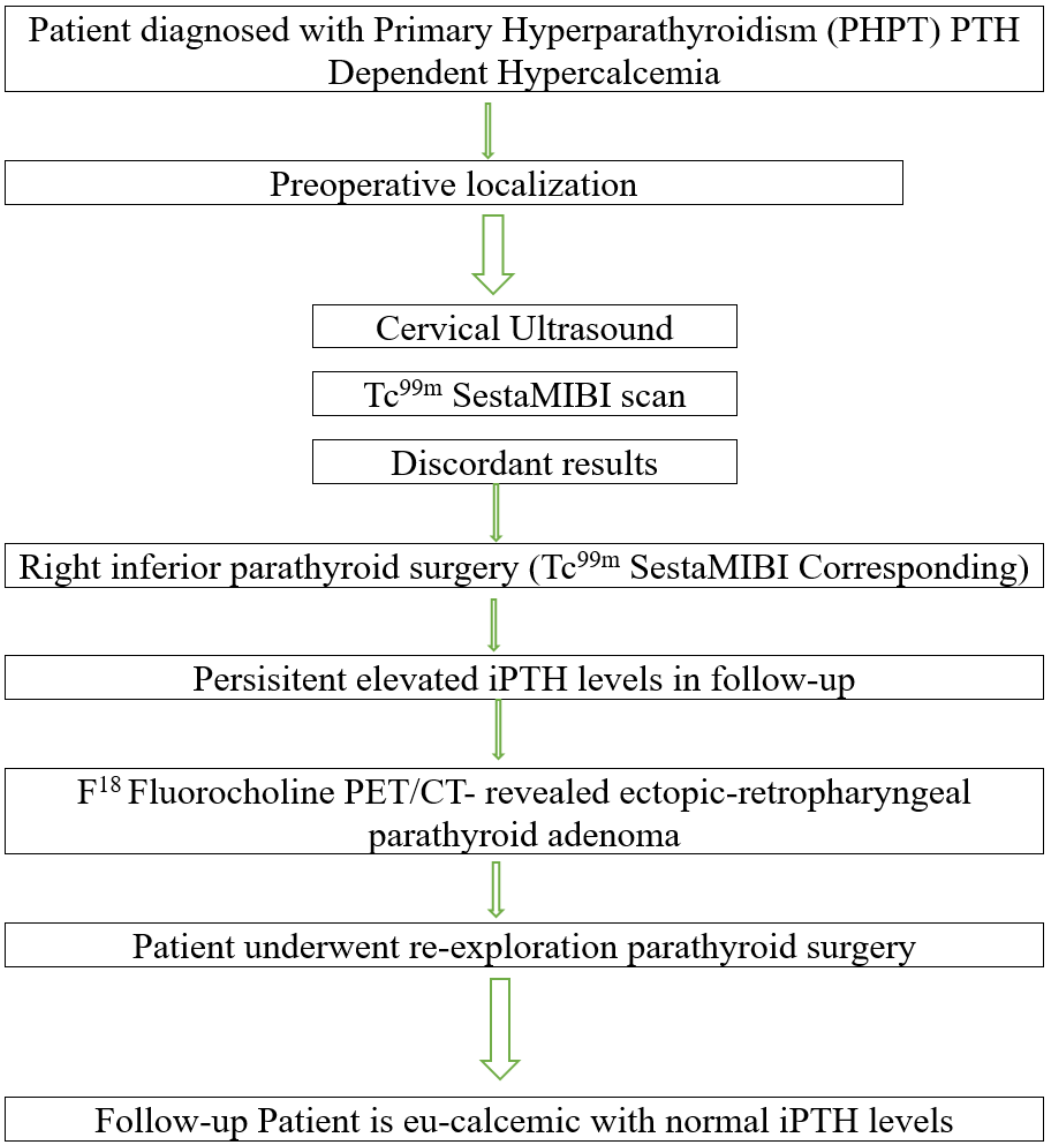

Figure 1: Flowchart of the process followed.

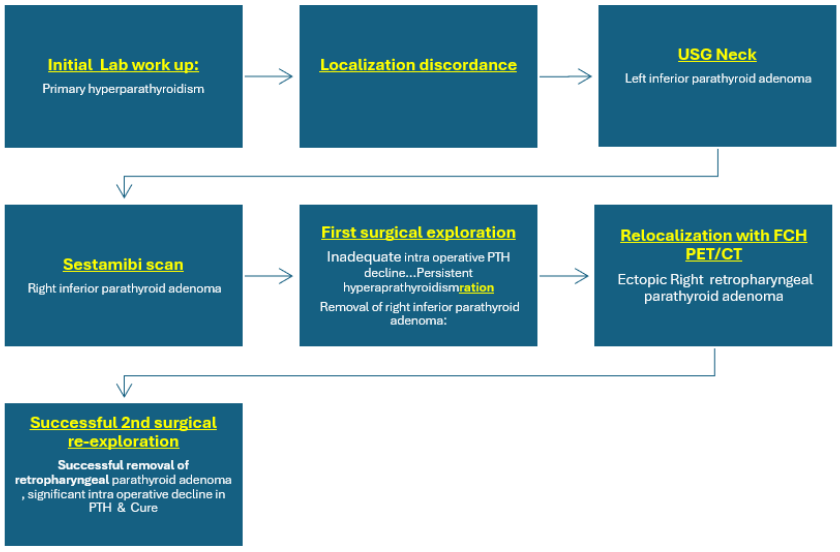

Figure 2: Flowchart of the studies done.

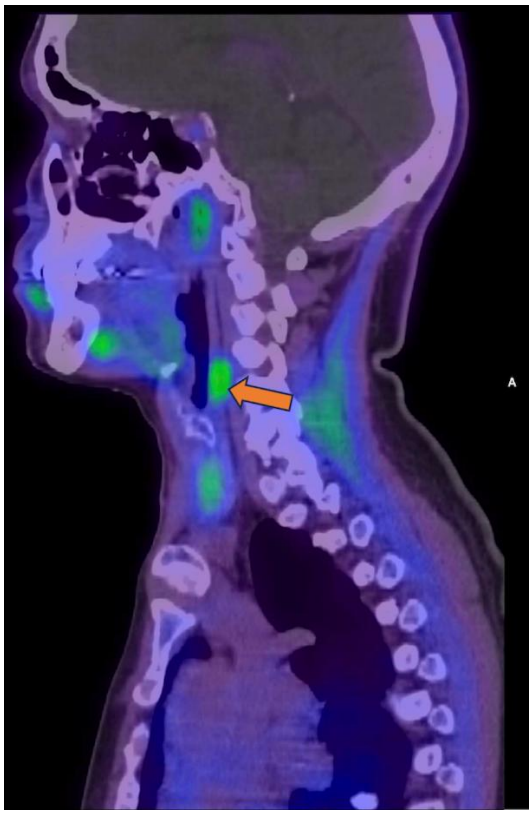

Figure 3: Sagittal fused PET/CT image of the neck demonstrating focal radiotracer uptake in the posterior cervical region, indicated by the orange arrow, corresponding to the suspected parathyroid lesion.

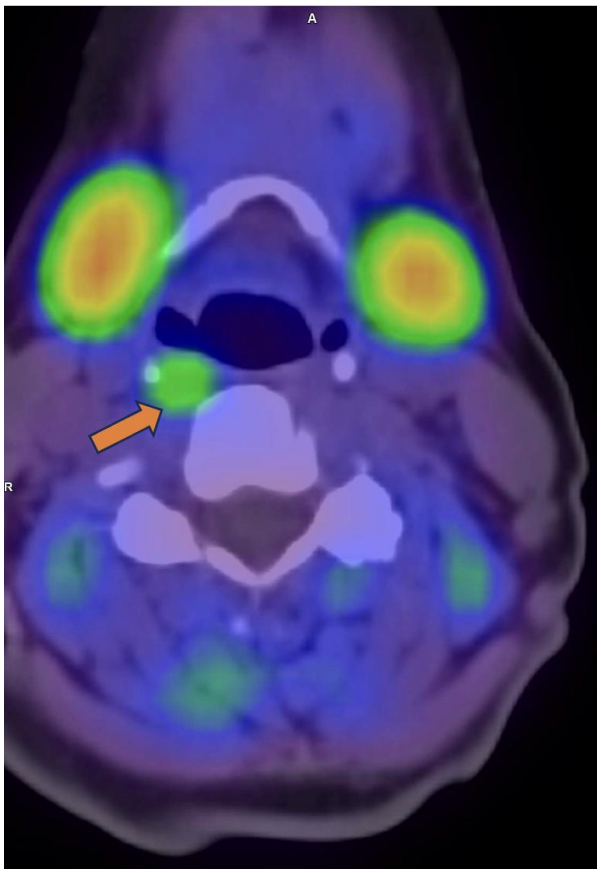

Figure 4: Axial fused PET/CT image of the neck demonstrating focal radiotracer uptake in the right posterior cervical/paravertebral region, indicated by the orange arrow, corresponding to the suspected parathyroid lesion.

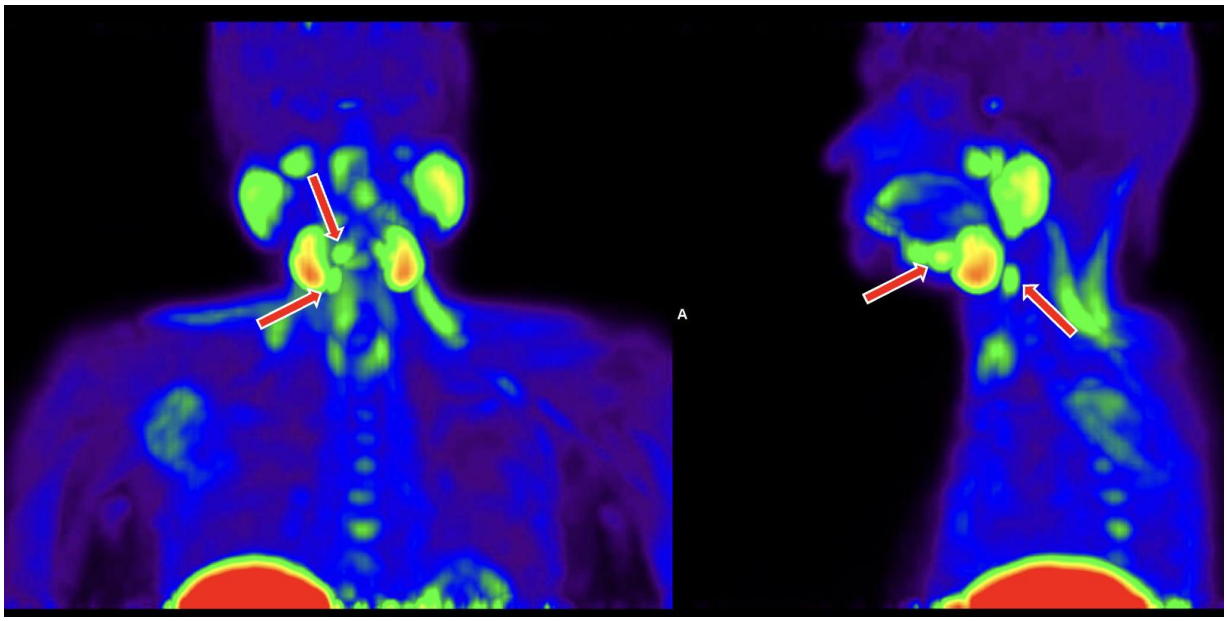

Figure 5: Anterior and lateral planar PET images demonstrating focal radiotracer uptake in the cervical region, indicated by the red arrows, consistent with parathyroid tissue localization.

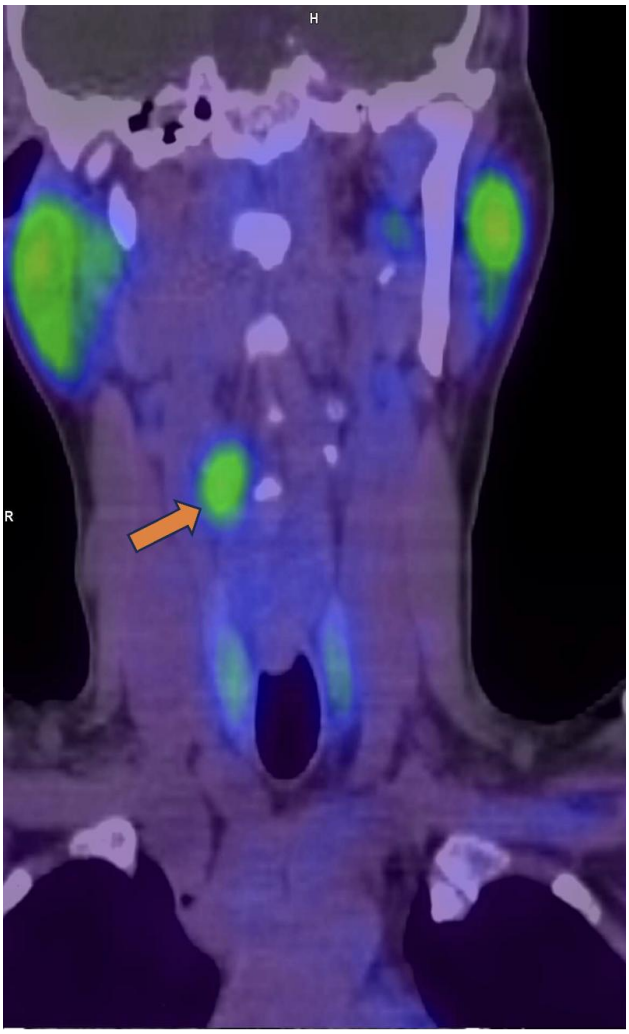

Figure 6: Coronal fused PET/CT image demonstrating focal radiotracer uptake in the lower cervical region, indicated by the orange arrow, corresponding to the suspected parathyroid lesion.

Discussion

Primary hyperparathyroidism pre-operative localization is a difficult task. The usual standard is to obtain concordant imaging by two techniques whenever possible. Hence the parathyroid imaging guidelines suggest imaging by two methods to improve the sensitivity of localization of the lesion [6]. Bilateral neck exploration is considered the gold standard for detection for parathyroid glands localization in primary hyperparathyroidism [2]. The cervical Ultrasonography (cUS) is cheapest and easily available tool. The Cervical ultrasound is having sensitivity of 16.2 % in double adenoma and 34.9% in multi-gland disease as compared to 78.5% in single gland disease [7]. The reliability of positive ultrasonographic imaging was high with 93.2% high positive predictive value [8]. The ability of the ultrasonography to correctly localize the enlarged parathyroid glands in primary hyperparathyroidism varies from 67-87% in patients without prior surgery [2].

Imaging cannot differentiate between parathyroid adenoma from carcinoma. Diagnosis is often accomplished after parathyroidectomy. The main role of imaging is to know the morphology including anatomical localization and the extent of parathyroid lesion and to locate the ectopic parathyroid gland [3].

In our case the pre-operative localization was not concordant on the two imaging modalities used, as cervical ultrasonography was suggestive of cystic lesion on left side of the neck and sesta MIBI localized the lesion on the right. The cervical ultrasound is anatomical imaging and SestaMIBI imaging is functional imaging. The cervical ultrasound is having a pooled sensitivity of 76.1 % and SestaMIBI is having a pooled sensitivity of 78.9% [8]. In patients with parathyroid reoperation the SestaMIBI is having higher sensitivity as compared to cervical ultrasound [9]. After careful consideration, we thought that the thyroid lesion might be an benign incidental cystic nodule. Considering the higher sensitivity of SestaMIBI we went ahead with right inferior parathyroid surgery [2,8]. The close follow up after parathyroid surgery helped us to detect the persistent elevated iPTH levels and intrigued us to search for another lesion. While we were thinking about persistence of hyperparathyroidism, we did Flurocholine PET/CT for this patient to look for any residual or ectopic tumor source. Fluorocholine PET has shown higher sensitivity to detect parathyroid lesion in re-operation cases as well as in pre-operative evaluation as compared to cervical ultrasound and SestaMIBI [9-11]. Fluorocholine PET/CT revealed retropharyngeal parathyroid lesion. Patient underwent second surgery and during this the intraoperative iPTH levels were recorded to be in the lower half of normal range. Subsequently patient showed normal iPTH and calcium levels during follow-up and is in remission now. The remission rate in cases of PHPT is 95% in experienced surgeons' hand, whereas the recurrence rates vary and increases in recent studies [12].

Conclusion

Our cases illustrate the routine discordance between imaging modalities for primary hyperparathyroidism. Flurocholine PET/CT is effective functional imaging modality, especially in cases of recurrence of PHPT. For routine pre-operative evaluation of cases with PHPT combination of cervical ultrasound and SestaMIBI improves localization sensitivity. Close monitoring of operated primary hyperparathyroidism cases should be done to pick up early recurrence.

Conflict of Interest

The authors declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

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Data Availability Statement

The data supporting the findings of this study are available from the corresponding author upon reasonable request.

Ethical Statement

The project did not meet the definition of human subject research under the preview of the IRB according to federal regulations and therefore was exempt.

Informed Consent Statement

Informed consent was obtained from all participants included in the study.

Authors' Contributions

All authors contributed equally to this paper.

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