CASE REPORT

Retropharyngeal Ectopic Parathyroid Adenoma: A Case Report with Review of Literature

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Abstract

Retropharyngeal ectopic parathyroid adenoma (PTA) is a rare entity. We present a case report of a patient with primary hyperparathyroidism (PHPT) who underwent dissection of the retropharyngeal space for excision of the PTA. It is imperative for an endocrine surgeon to be familiar with potential ectopic locations of PTA, especially when the results of routine first-line radiographic studies are equivocal. This awareness will avoid unsuccessful neck explorations and prepare the surgeon for a minimally invasive approach to excise the PTA. This case highlights the retropharyngeal space as an important ectopic site for PTA, underscores the limitations of ultrasound, and emphasizes the importance of newer diagnostic modalities such as technetium 99m sestamibi (MIBI) and 4D-computed tomography (4DCT) scans in easy preoperative identification of the location of ectopic PTAs.

Keywords: Adenoma, Case report, Ectopic, Hyperparathyroidism, Parathyroid.

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Introduction

For individuals with symptomatic primary hyperparathyroidism (PHPT) and for those with asymptomatic PHPT who meet the National Institutes of Health criteria, the advised treatment is surgical excision of the parathyroid adenoma (PTA).[1] The primary aim of preoperative imaging in PHPT is to accurately identify the location of a single PTA or to detect multiple gland disease.[2] Accurate preoperative localization of the PTA enables a minimally invasive parathyroidectomy (MIP) approach.[2] If localization is unsuccessful, a standard bilateral neck exploration is conducted, followed by either complete or partial parathyroidectomy. Serial measurement of intraoperative serum parathormone (PTH) levels are frequently utilized to evaluate the adequacy and success of surgical removal.[3]

The majority of PTAs are found near the thyroid gland, with ectopic PTA occurring in about 6.3–16% of patients undergoing surgery for PHPT. Identifying ectopic PTA accurately with preoperative imaging is essential to determining the best surgical method and incision location, reducing the risk of surgical failure. Tc-99m-sestamibi single photon emission computed tomography (SPECT)/computed tomography (CT) is commonly employed to locate PTA, including cases that are ectopic cases. Ultrasound (USG) is typically used alongside other imaging modalities, especially for evaluating any coexisting thyroid conditions. Problem-solving imaging techniques, such as contrast-enhanced 4D-computed tomography (4DCT) or contrast-enhanced magnetic resonance imaging (MRI), are often used as well. However, despite these imaging options, differentiating PTA from similar structures like thyroid nodules or lymph nodes can still be difficult before surgery.

We present a case of PHPT with ectopic retropharyngeal PTA, initially misdiagnosed on USG as a thyroid nodule or lymph node. The correct diagnosis was later made using Tc-99m-sestamibi SPECT/CT. She was successfully treated with MIP.

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Case Presentation

A female in her late 30s presented with recurrent renal stones for the last 12 years, with generalized body aches for the last 2 years. She experienced multiple episodes of pyelonephritis and cystitis, leading to several hospital admissions. She was diabetic and hypertensive for the last 10 years on regular medication.

Investigations

On routine investigations, she was found to have hypercalcemia (10.9 mg/dL) following which intact parathyroid hormone (iPTH) level was assessed, which was found to be elevated (142 pg/mL) Preoperative USG was not successful in differentiating the PTA from a thyroid nodule and lymph node. A sestamibi scan revealed an area of uptake close to the base of the tongue, indicative of an ectopic PTA (Fig. 1). A 4D-CT scan was done to know the exact location of the ectopic parathyroid gland, so a minimally invasive surgical approach can be taken in this case (Fig. 2). A preoperative

diagnosis of PHPT with ectopic retropharyngeal PTA was made with the help of a Tc-99 Sestamibi scan and a 4D-CT scan of the neck.

Treatment

The patient underwent a focused parathyroidectomy. Intraoperatively, a 2.5 × 1.5 cm retropharyngeal PTA was identified, extending from hyoid bone to the superior border of thyroid cartilage on the right side and weighing 2.9 gm. Intraoperative image of the specimen is shown (Fig. 3). The specimen photo is shown separately (Fig. 4). Immediate postoperative period iPTH level dropped to <3.6 pg/mL and total calcium to 8.6 mg/dL. On the second day of surgery, the patient did not have any signs or symptoms of hypocalcemia and was discharged from the hospital.

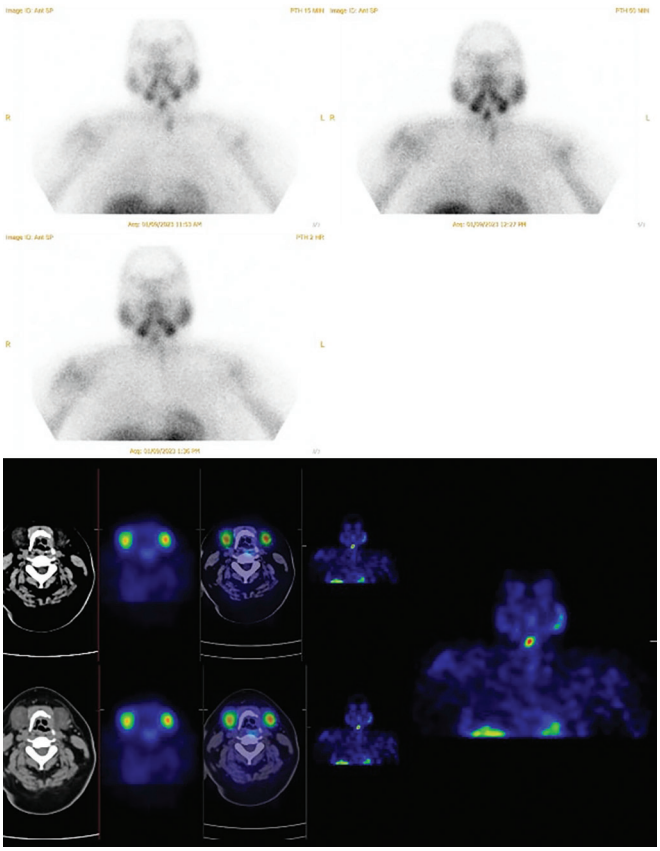

Fig. 1: Tc-99 Sestamibi scan demonstrating a focus of uptake near the base of the tongue, suggestive of an ectopic parathyroid adenoma

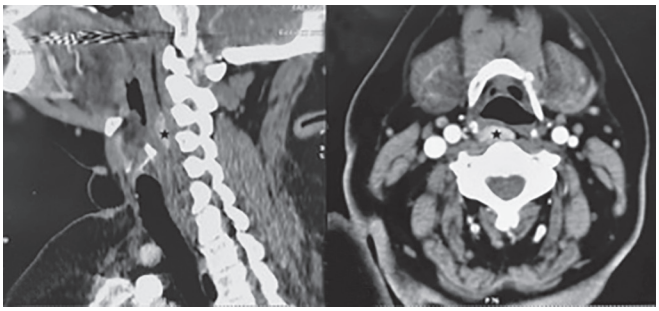

Fig. 2: 4DCT scan of the neck showing retropharyngeal parathyroid adenoma extending from hyoid bone to the superior border of the thyroid (marked in *)

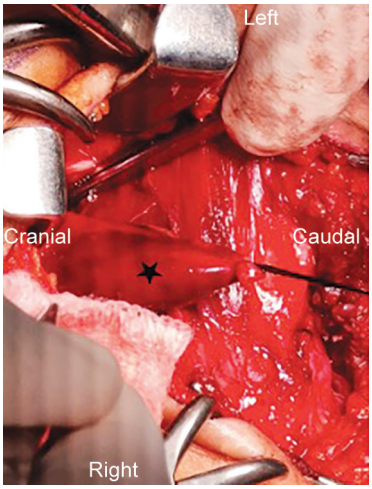

Fig. 3: Intraoperative image showing the dissection of the ectopic parathyroid gland from the retropharyngeal area (parathyroid gland marked in*)

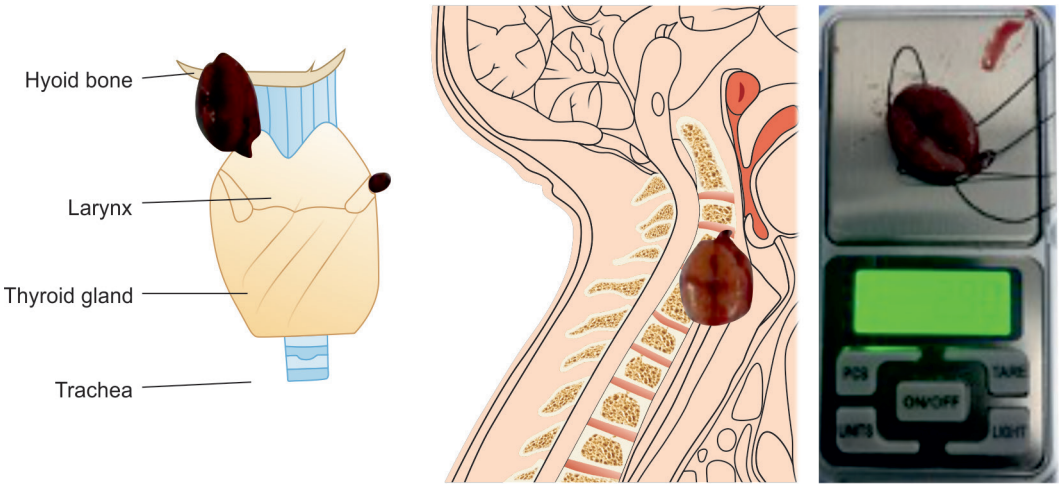

Fig. 4: Figure showing the excised ectopic parathyroid gland

On final pathological evaluation, the lesion measured 3 × 1.5 × 1.2 cm and weighed 2.82 grams. Multiple sections show features of PTA composed of chief cells with normal parathyroid in the periphery. The diagnosis of PTA was subsequently confirmed on histopathology.

Outcome and Follow-up

At 3 months' follow-up, the patient had a normal iPTH and calcium value with no symptoms or signs of hypo or hypercalcemia.

Discussion

A solitary PTA is present in 85% of cases with PHPT.[4,5] Most frequently, PTAs are seen adjacent to the inferior or posterior portion of the thyroid gland. Ectopic PTAs have an incidence between 2 and 22%, and they are characterized by their location outside the juxta-thyroid position.[6–10] Any point along their embryologic lineage can be the site of ectopic PTAs.[8] Thymus, retro-esophageal, intra-thyroidal, mediastinal, and inside the carotid sheath are the most typical sites for ectopic PTA.[7]

Even more so in patients who are being considered for a second operation, the retropharyngeal region represents an essential but unusual ectopic location of PTA. Gallagher and colleagues, in their cohort of patients with sporadic PHPT, reported a failure rate of 17% for initial surgery in retropharyngeal glands, in contrast to a 3.1% failure rate for eutopic gland.[11] The study found that retropharyngeal glands were four times more frequently identified during reoperations, highlighting the retropharyngeal area as a potential "blind spot" for locating PTAs. The first line imaging method used for preoperative localization of PTA is Tc-99m-sestamibi SPECT/CT with reported sensitivity and accuracy of 90 and 97.2% respectively. However, the preferred imaging technique may vary depending on the institution.[12,13] While the specificity of Tc-99m-sestamibi SPECT/CT is not often discussed, false positives can arise from uptake in lymph nodes and thyroid nodules that could be reactive, granulomatous, or metastatic.[14] The finding of the "polar vessel sign" is helpful in distinguishing PTA from a reactive lymph node, which typically exhibits a fatty hilum in the majority of cases.[15] Two-thirds of solitary PTA cases have been observed to have the "polar vessel sign," which is characterized by an enlarged branch of an artery or an early draining vein originating from the PTA's poles and is more common with larger-sized PTA.[15] Ultrasound has limitations in identifying a retropharyngeal PTA due to a lack of an adequate acoustic window. The negative result on Tc-99m-sestamibi planar imaging is likely due to the deeper location of the ectopic PTA, and the lower sensitivity of planar imaging has been documented. A careful examination of a patient's laboratory results and knowledge of the imaging appearance of PTA on 4D-CT and MRI can also help identify asymptomatic PTA's, whether the more common eutopic or the uncommon ectopic retropharyngeal, as in our case. This is because a sizable portion of PHPT is asymptomatic.

Tc-99 Sestamibi SPECT is frequently used to localize PTAs. When this scan is negative in cases of PHPT, 4D-CT or MRI are often the next steps. In our case, the retropharyngeal PTA showed uptake on a Tc-99 Sestamibi scan. 4D-CT imaging includes three spatial planes, while the fourth dimension represents perfusion data obtained from the non-contrast, arterial, and delayed venous phases.[16] Parathyroid adenomas characteristically display rapid contrast washout during the delayed venous phase and appear hyper-enhanced in the arterial phase.

In our institution, we often utilize sonography and sestamibi scintigraphy to locate PTAs. Besides detecting parathyroids, ultrasonography can inform clinicians of concurrent thyroid conditions, making it an ideal first-line approach to reduce additional radiation exposure and the costs associated with 4D-CT. If these initial modalities do not locate the lesion, a 4D-CT is then employed. Magnetic resonance imaging is also sometimes used for parathyroid localization, providing a radiation-free alternative with sensitivity comparable to sonography and sestamibi.[17,18] Magnetic resonance imaging may improve detection with varied T1 and T2 signals, and PTAs may show post-contrast enhancement on MRI.[18] Nonetheless, cervical lymph nodes might exhibit comparable signal features and patterns of enhancement, and respiratory motion can further complicate MRI imaging accuracy.

Over the last decade, minimally invasive surgical methods have gained popularity for the excision of PTAs, largely due to advanced imaging methods that enhance preoperative localization. Minimally invasive parathyroidectomy, typically conducted via a small midline incision and is frequently used for small and well-localized adenomas, and has mostly replaced conventional cervical four-gland exploration for single-gland disease.[18] Surgeons who are aware of retropharyngeal ectopic PTAs are better equipped to avoid unsuccessful neck explorations and perform minimally invasive excision of PTAs irrespective of their location.

Take-home Messages

- This case is among the few documented in the literature where a retropharyngeal PTA was surgically confirmed and cured an ectopic site known for its higher rate of initial surgical failure.
- It demonstrates the effectiveness of Tc-99m-Sestamibi SPECT/CT in accurately identifying ectopic retropharyngeal PTA, which enabled a successful, targeted, and minimally invasive parathyroidectomy.
- This case also highlights the advantages and limitations of the various preoperative imaging modalities and emphasizes the importance of thorough image review alongside relevant clinical details to avoid overlooked or misinterpreted findings.

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