An Unusual Ectopic Parathyroid Adenoma Location: Pyriform Sinus

Case Report

Enver Can Öncül[1], Gökçen Güngör Semiz[2], Erkan Derebek[3], Ersoy Doğan[1]

[1]Dokuz Eylül University Faculty of Medicine, Department of Otorhinolaryngology, İzmir, Türkiye
[2]Dokuz Eylül University Faculty of Medicine, Department of Endocrinology and Metabolism, İzmir, Türkiye
[3]Dokuz Eylül University Faculty of Medicine, Department of Nuclear Medicine, İzmir, Türkiye

Abstract

Primary hyperparathyroidism (pHPT) is caused by a single or multiple parathyroid adenomas in 85% of cases, by parathyroid hyperplasia in 15%, and by parathyroid carcinomas in less than 1%. Due to the embryological development characteristics of parathyroid tissues, ectopic parathyroid adenomas can be found in various locations. Although the incidence of pHPT varies, it is not considered a rare disease. Some unusual locations of ectopic parathyroid adenomas can make it difficult for clinicians and surgeons to detect them. For this reason, it is important to understand embryology thoroughly and to use complementary preoperative imaging methods. The detection and accurate localization of adenomas are important to avoid recurrent surgeries and to increase surgical success. We aimed to contribute to the literature by presenting a case of an adenoma located in the right pyriform sinus.

Keywords: Head and neck surgery, hyperparathyroidism, parathormone, parathyroid adenomas, pyriform sinus, case report

ORCID IDs of the authors:
E.C.Ö. 0000-0002-2058-7369
G.G.S. 0000-0002-8915-9178
E.D. 0000-0002-5161-7655
E.D. 0000-0003-0539-4232

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Corresponding Author:
Enver Can Öncül, MD;
envercanoncul@hotmail.com

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Introduction

The incidence of primary hyperparathyroidism (pHPT) is relatively common, with estimates ranging widely between 0.4 and 82 per 100,000 population. The incidence of pHPT has increased significantly in countries where routine biochemical screening for parathyroid hormone (PTH) levels has been implemented (1). It can cause a wide range of clinical symptoms. Although most patients are clinically asymptomatic, some may present with non-specific complaints such as fatigue, cognitive impairment, and mild depression. These symptoms result from prolonged elevated levels of PTH and the associated hypercalcemia.

In affected individuals, complications may include renal disorders such as nephrolithiasis and renal failure; gastrointestinal issues such as nausea, vomiting, peptic ulcers, and even pancreatitis; musculoskeletal symptoms such as bone pain, muscle weakness, pathological fractures, and osteitis fibrosa cystica; and neuropsychiatric manifestations such as lethargy, psychosis, and coma (2).

There are various underlying pathologies that can cause pHPT. Approximately 85% of cases are due to single or multiple parathyroid adenomas, 15% are attributed to parathyroid hyperplasia, and less than 1% are caused by parathyroid carcinoma.

Although parathyroid adenomas can occur at any age, they are most commonly diagnosed in the fifth and sixth decades of life. While some studies suggest that adenomas of the inferior parathyroid glands are more frequent than those of the superior glands, contradictory evidence also exists in the literature (3).

We aimed to contribute to the literature by presenting a case of an ectopic parathyroid adenoma located in the pyriform sinus, an extremely rare location.

Case Presentation

A 53-year-old female patient presented with complaints of excessive water intake for the past two years, bone pain in her right ankle, stiffness in her arms and hands, and generalized weakness. The patient had no history of pathological bone fractures, nephrolithiasis, gastrointestinal problems, or a family history of hyperparathyroidism. Flexible endoscopy revealed a raised lesion on the medial wall of the right pyriform sinus. Other ear nose and throat and systemic examinations were unremarkable.

Serum total calcium level was 11.37 mg/dL (normal: 8.8-10.6 mg/dL), albumin level was 4.25 g/dL (reference range: 3.5-5.2 g/dL), and phosphorus level was 2.38 mg/dL (reference: 2.7-4.5 mg/dL). The serum parathormone (PTH) level for differential diagnosis was markedly elevated at 516.9 pg/mL (reference: 14-72 pg/mL). Neck ultrasonography (USG) did not reveal any lesions compatible with parathyroid adenoma in either eutopic or ectopic locations.

In Technetium 99m sestamibi (99mTc-sestamibi) single-photon emission computed tomography (SPECT/CT) imaging, focal tracer uptake was observed at the level of the superior right thyroid lobe, showing retention suggestive of an ectopic parathyroid adenoma, consistent with the lesion seen on CT (Figure 1). Four-dimensional CT (4D-CT) revealed a lesion measuring approximately 10×10×14 mm in the right pyriform sinus, consistent with a parathyroid adenoma (Figure 2a, b, c).

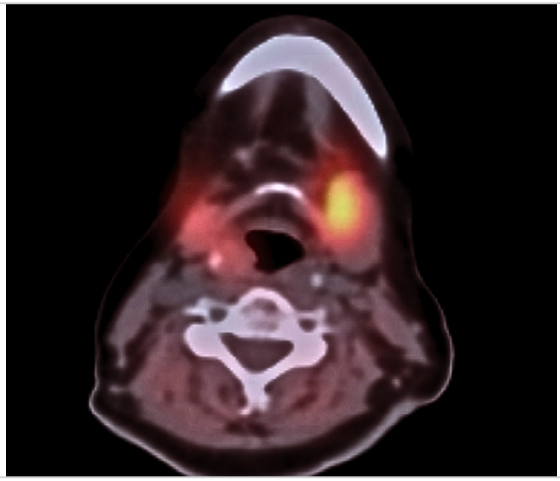

Figure 1. SPECT/CT imaging demonstrates focal radiotracer uptake in the right pyriform sinus, consistent with an ectopic parathyroid adenoma
SPECT/CT: Single-photon emission computed tomography

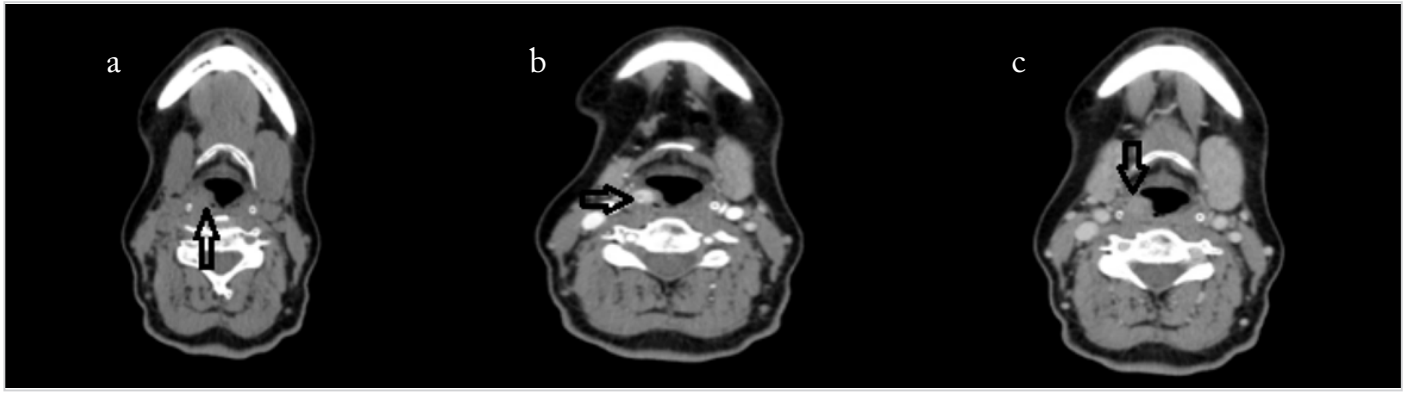

Figure 2. Axial sections of neck computed tomography showing a lesion consistent with parathyroid adenoma in the right pyriform sinus: pre-contrast, early post-contrast, and delayed post-contrast phases

The patient underwent direct laryngoscopy under general anesthesia. During the procedure, a capsulated mass measuring approximately 2 cm was observed on the medial wall of the right pyriform sinus (Figure 3). The mass was excised using microscissors and laryngeal cautery. The intraoperative rapid PTH test showed that the PTH value, 406.5 pg/mL before excision, had decreased to 31.3 pg/mL approximately 15 minutes after excision. Postoperatively, both PTH and serum calcium levels returned to within normal limits. The histopathological examination of the surgical specimen confirmed the diagnosis of parathyroid adenoma (Figure 4). The patient was discharged uneventfully. At the last follow-up, conducted 18 months postoperatively, no recurrence or pathology was observed. Informed consent was obtained from the patient for publication.

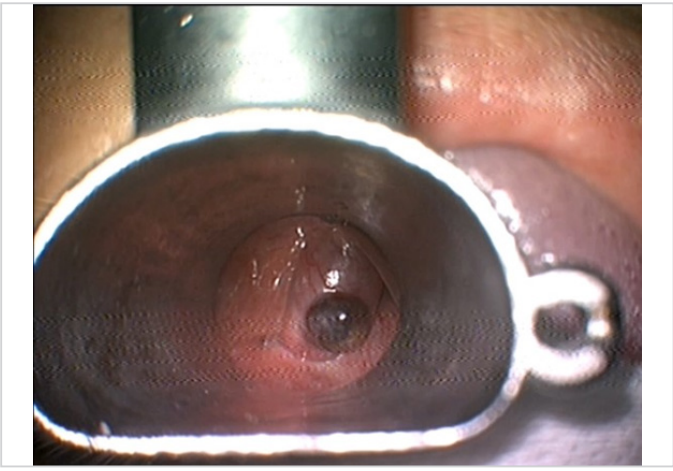

Figure 3. Intraoperative direct laryngoscopy image showing a well-defined lesion on the medial wall of the right pyriform sinus, suggestive of an ectopic parathyroid adenoma

Figure 4. Image of the mass excised during surgery

Discussion

The ectopic location of parathyroid adenomas has been attributed to their abnormal migration during embryogenesis. Five pairs of pharyngeal pouches are formed when the embryo is approximately 26 days old. Parathyroid glands begin to develop during the fifth week of intrauterine life. The inferior glands originate from the third pharyngeal pouch, while the superior glands arise from the fourth pharyngeal pouch, becoming histologically visible by the 14th week. The fact that the fourth pharyngeal pouch also contributes to the development of the pyriform sinus explains the ectopic location observed in this case (4). Several previous case reports have described parathyroid adenomas located in this rare anatomical region (5-9).

Currently, imaging methods such as preoperative USG, 99mTc-sestamibi scintigraphy (MIBI), and 4D-CT are available to determine the location of parathyroid adenomas.

The typical ultrasonographic appearance of a parathyroid adenoma is an oval-shaped, hypoechoic nodule closely associated with the thyroid gland. However, ectopic parathyroid adenomas may be missed on USG due to factors such as operator experience, interpretive error, or technical limitations like the absence of an acoustic window. The sensitivity and positive predictive value (PPV) of USG have been reported as 76.1% and 93.2%, respectively (10). In our case, USG was used as the initial imaging modality; however, no suspicious lesion was detected. This highlights the importance of considering alternative imaging modalities in cases of suspected parathyroid adenoma when USG findings are negative.

99mTc-sestamibi SPECT has pooled sensitivity and PPV rates of 78.9% and 90.7%, respectively (10). As demonstrated in our case, we consider this imaging technique to be both complementary and reliable.

The 4D-CT is another widely used imaging tool. With this technique, abnormal parathyroid glands as small as 1×6 mm can be identified. It is particularly useful in detecting adenomas that are missed by other imaging methods or in cases of unsuccessful prior neck explorations. However, the radiation exposure associated with 4D-CT is a notable disadvantage compared to standard CT. The overall sensitivity and PPV of contrast-enhanced neck CT, including the 4D variant, have been reported as 89.4% and 93.5%, respectively (10).

In our clinical practice, if the findings of USG and SPECT/PET are consistent, we consider the localization to be adequately established and typically do not proceed with 4D-CT. However, when discrepancies exist between USG and SPECT/PET findings, we believe that 4D-CT is useful both for precise localization and for planning an accurate surgical approach.

Conclusion

Some uncommon locations of ectopic parathyroid adenomas may pose significant challenges for clinicians and surgeons in detecting the adenoma. For this reason, a thorough understanding of parathyroid embryology and the use of complementary preoperative imaging modalities are essential. The detection and precise localization of adenomas play a critical role in preventing recurrent surgeries and in improving surgical outcomes.

Ethics

Informed Consent: Informed consent was obtained from the patient for publication.

Footnotes

Authorship Contributions

Surgical and Medical Practices: E.C.Ö., E.D., Concept: E.C.Ö., E.D., Design: E.C.Ö., E.D., Data Collection and/or Processing: E.C.Ö., G.G.S., E.D., E.D., Analysis and/or Interpretation: E.C.Ö., G.G.S., E.D., E.D., Literature Search: E.C.Ö., E.D., Writing: E.C.Ö., E.D.

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Main Points

- Parathyroid adenomas are an important pathology that can lead to various gastrointestinal, musculoskeletal, renal, and neuropsychiatric complications.
- Due to their embryological development, parathyroid adenomas may be located in unusual anatomical sites.
- Preoperative imaging particularly ultrasound, four-dimensional computed tomography, and Technetium 99m sestamibi single-photon emission computed tomography plays a crucial role in determining the exact location of the adenoma.

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