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CASE REPORT

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Undescended superior parathyroid: A case report

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Abstract

Aberrant migration of parathyroid glands from their embryologic origin may result in undescended parathyroid glands. We present a case of an ectopic parathyroid adenoma at the level of the pyriform sinus. A 41-year-old female was evaluated for primary hyperparathyroidism. Following non-localizing ultrasound and planar sestamibi imaging, the patient underwent SPECT/CT and 4-D computed tomography demonstrating evidence of an ectopic parathyroid adenoma. The surgical approach was modified based on the location. Following extirpation, PTH fell from 80 to 16 pg/mL, and the 15-min post-excision level remained stable at 14pg/mL, indicating a biochemical cure. While rare, undescended parathyroid adenoma should be considered when preoperative imaging fails to identify a target adenoma or after unsuccessful surgery. The combined use of 99m Tc-MBI or 4D CT and other anatomical scans may improve diagnostic accuracy. Due to the potential need to perform a second incision to conduct a four-gland exploration, preoperative patient discussion regarding surgical risks may differ from that of a standard parathyroidectomy.

KEYWORDS

ectopic parathyroid, parathyroid adenoma, parathyroidectomy, primary hyperparathyroidism, undescended parathyroid gland

1 | INTRODUCTION

Parathyroid glands normally descend from their embryologic origin in the pharyngeal pouches to their final locations in the central neck. However, aberrant migrations can lead to one or more parathyroid glands being in nonanatomic locations, including retroesophageal space, thymus, mediastinum, or within the thyroid gland. Rarely they fail to migrate, resulting in an undescended gland. Adenomas arising from these locations should be considered when initial preoperative imaging fails to identify a target adenoma or after unsuccessful surgery. Here we present a case of an ectopic parathyroid adenoma arising at the level of the pyriform sinus.

2 | CASE REPORT

A 41-year-old female presented to our institution for evaluation of primary hyperparathyroidism. She had a 7-year history of hypercalcemia, with levels up to 11.5 mg/dL (reference range: 8.7–10.2 mg/dL) and PTH levels up to 101 pg/mL (reference range: 10–73 pg/mL). She has no pertinent family history and is a former smoker (0.25 ppd, 5 pack-years). Initially, she underwent an ultrasound (US) of the neck and ^{99m}Tc-MIBI single photon emission computed tomography (SPECT) imaging, which failed to identify any suspicious lesions or localizing parathyroid adenomas. A slight asymmetric tracing could be visualized just caudal to the submandibular gland at the level

of the piriform sinus in the SPECT imaging (Figure 1). However, it was difficult to confirm the presence of parathyroid adenoma in this imaging modality, given its proximity to the high uptakes in the submandibular gland. Furthermore, this location is a frequent site for enlarged or normal cervical lymph nodes, further complicating the differentiation from a parathyroid adenoma using ultrasonography. Subsequent SPECT/CT (Figure 1) and 4-D computed tomography (CT) (Figure 2) demonstrated evidence of a 1.3 cm lesion superior to the thyroid at the level of the pyriform sinus on the left side, likely representative of an ectopic or undescended parathyroid adenoma. Given its unusual location patient was preoperatively counseled on the risk of injury to the hypoglossal nerve, marginal mandibular nerve, and the possible need for a second incision for four gland exploration if the candidate lesion was not an adenoma.

On the day of surgery, preoperative PTH level was 80 pg/mL. In the operating room, an upper transcervical incision was performed based on its anatomical location seen on the CT imaging caudal to the submandibular gland and superior to the thyroid at the level of the piriform sinus. An US was performed to confirm the location of the lesion preoperatively. The subplatysmal flap was raised superiorly and inferiorly to expose the inferior aspect of the submandibular gland. Then the investing fascia was incised just caudal to the submandibular gland, which was retracted superiorly to reveal the posterior belly of the digastric muscle. The digastric muscle was dissected along its anterior face and subsequently retracted superiorly to identify the hypoglossal nerve and the internal jugular vein. A crossing facial vein was encountered that required ligation to gain appropriate access. An enlarged and undescended parathyroid measuring 1.5 × 1.5 cm located deep and slightly inferior to the anterior belly of the digastric muscle, anterior and slightly medial to the carotid artery, was identified and carefully resected (Figure 3, Figure 4).

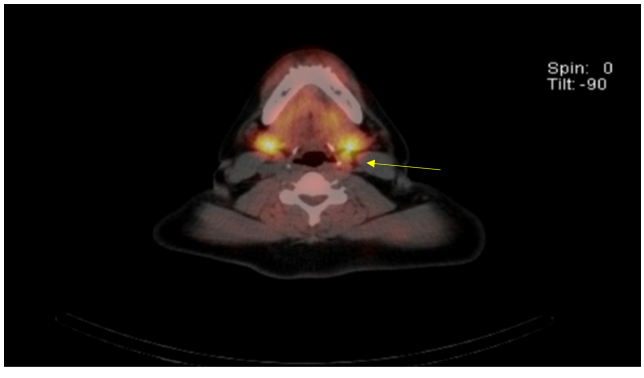

FIGURE 1 Sestamibi Single Photon Emission Computed Tomographic (SPECT/CT) Study. Yellow arrow points to the candidate lesion.

10 min following the excision of the adenoma, PTH fell to 16 pg/mL and at 15 min post-excision remained stable at 14 pg/mL indicating biochemical cure.

3 | DISCUSSION

Undescended parathyroid glands are typically defined as those located at or above the carotid bifurcation.[1] This classification of parathyroid glands only constitutes 2%–7% of parathyroid adenomas, making it the least common form of parathyroid glands found in ectopic locations.[2] While rare, undescended parathyroid adenoma should be considered when initial preoperative imaging fails to identify a target adenoma or after unsuccessful parathyroid surgery.

Preoperative localization studies are critical in the identification of possible undescended parathyroid adenomas. Imaging modalities typically include high

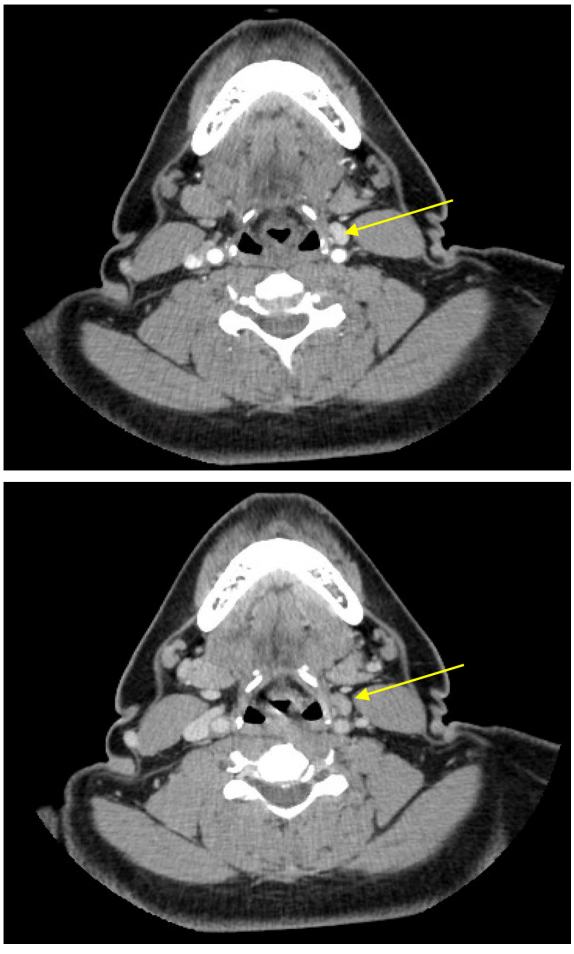

FIGURE 2 Preoperative 4D-CT scan. Yellow arrows point to the candidate lesion. The top image reveals an arterial hyperenhancement of the candidate lesion. The bottom image shows rapid washout on the venous phase of the candidate lesion.

FIGURE 3 Removed ectopic parathyroid gland with the illustration of relevant anatomical structures.

FIGURE 4 Intraoperative image of surgical incision site.

resolution neck US, SPECT, parathyroid scintigraphy, CT and/or magnetic resonance imaging (MRI). In a meta-analysis of 1276 patients, Wong et al. demonstrate a sensitivity of 86% [Confidence Interval: 0.81–9.90] for ^{99m}Tc-sestamibi SPECT/CT in identifying ectopic parathyroid adenomas, which is superior to the sensitivity of SPECT and planar imaging modalities alone.[3] The addition of anatomical imaging can further enhance the diagnostic localization of the candidate lesion. In a review of 656 patients at a single institution, Zerizer et al. found that combining ^{99m}Tc-MIBI with anatomical scans (CT or MRI) significantly improved diagnostic accuracy (improving sensitivity and specificity to 100%).[4] Anecdotally, the localization of parathyroid adenomas can be complicated by false positive signals generated by thyroid nodules as well as the salivary glands, as was the case for our undescended parathyroid. Additional imaging modalities, such as CT neck with contrast or MRI can be helpful to pinpoint the location and distinguish it from the neighboring structure as it did in this case. A recent review article demonstrated that MRI had both a sensitivity and specificity of up to 97% in the detection of parathyroid adenomas.[5] Common MRI traits of adenomas can include elongated morphology, T2 fat saturation hyperintensity, and strong enhancement T1 post-contrast.[5]

Four-dimensional CT has shown promise for the detection of ectopic parathyroid glands, but has the drawback of a greater radiation exposure compared to the sestamibi SPECT modality.[6] Four-dimensional CT specifically used for localization of ectopic parathyroids has not been clearly defined in the literature, however, it demonstrates a higher sensitivity (82%) and specificity (92%) compared to other imaging modalities, suggesting its benefit as a useful adjunct to localizing ectopic parathyroid adenomas.[7] While the protocols can vary, during 4-D CT, imaging series are captured using the intravenous contrast in three phases—non-contrasted, arterial, and delayed (venous). Different enhancement patterns in these three phases allow the characterization of parathyroid adenoma and differentiate the lesion from thyroid nodules or lymph nodes.[8] In our case, there was a distinct enhancement of the candidate lesion in the arterial phase with washout on the delayed or venous phase (Figure 2), suggesting the presence of parathyroid adenoma caudal to the left submandibular gland, superior and separate from the thyroid gland. Normal parathyroid glands were not visualized in this imaging modality.

Importantly, in preoperative planning, it is imperative to consider the potential need to perform a second incision in order to conduct a four-gland exploration in the case that the PTH did not decrease by >50% after the removal of the adenoma. The unique location of the surgical incision employed for this patient lends itself to potential surgical complications that are otherwise uncommon in standard parathyroidectomies. Specifically, surgeons must consider the increased risk of injury to the hypoglossal and marginal mandibular nerves, and injuries to the carotid artery, internal jugular vein, and vagus nerve if dissection of the gland is required from these structures. Furthermore, the use of a two-incision surgical approach necessitates the surgeon to counsel patients on potentially managing two scars post-surgery. As such, it is important to note that preoperative discussions with patients regarding surgical risks may differ from that of a standard parathyroidectomy.

AUTHOR CONTRIBUTIONS

Elliott M. Sina: Conceptualization; data curation; investigation; methodology; project administration; resources; software; validation; visualization; writing – original draft; writing – review and editing. **Chihun J. Han:** Conceptualization; investigation; methodology; resources; supervision; writing – review and editing. **Elizabeth E. Cottrill:** Conceptualization; data curation; investigation; methodology; project administration; resources; supervision; writing – review and editing.

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DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

CONSENT

Written patient consent has been signed and collected in accordance with the journal's patient consent policy.

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