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CASE REPORT

Ectopic pyriform sinus parathyroid adenoma

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KEYWORDS
Adenoma;
Parathyroid;
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Ectopic

Summary
Objective: To describe the diagnosis and treatment of ectopic pyriform sinus parathyroid adenoma.
Material and methods: A 44-year-old woman presented with persistent primary hyperparathyroidism after previous failed cervical exploratory surgery.
Results: Diagnosis of ectopic pyriform sinus parathyroid adenoma was suggested by computed tomography and technetium-99 m sestamibi scintigraphy (99mTc-MIBI SPECT/CT). A submucosal tumor was identified under laryngoscopy and resected by endoscopic CO_2 laser. Histopathology confirmed the diagnosis of parathyroid adenoma.
Conclusions: Ectopic pyriform sinus locations are rare in parathyroid adenoma. 99mTc-MIBI SPECT/CT facilitates diagnosis, especially in case of previous failed neck exploration. Endoscopic CO_2 laser resection is the treatment of choice.

Introduction

Ectopic parathyroid adenoma is a significant cause of post-surgical persistent primary hyperparathyroidism (PHPT) [1].

Pre-operative assessment generally allows diagnosis of the ectopic gland, but there remains a non-negligible risk of false negatives, as not all parathyroid lesions take up sestamibi in the same way [2].

False positives on this examination are much rarer, often due to associated nodular thyroid pathology [3].

A pyriform sinus location is quite exceptional; there has been only one report of management by endoscopic CO_2 laser [4].

We report the case of a woman presenting with ectopic adenoma of the pyriform sinus, not diagnosed on initial scintigraphy, and treated, after failure of classic neck exploration, by endoscopic CO_2 laser.

Case report

A 44-year-old woman was referred for surgical management of PHPT. Two successive assessments found hypercalcemia and inadequate parathormonemia (ionized calcium and PTH:

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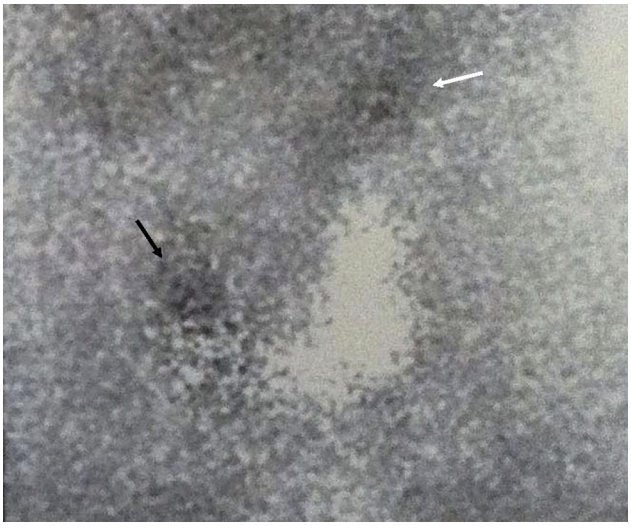

Figure 1 Weighted subtraction images after planar scintigraphy (^{99m}Tc/^{99m}Tc-MIBI). Hyperfixation at right parathyroid P4 (black arrow). NB: moderate fixation could be discerned on retrospective re-reading in the inferior part of the left submandibular fovea (white arrow), doubtless corresponding to the pyriform sinus adenoma.

1.48 mmol/L; 56 pg/L (< 38.8), and 1.36 mmol/L; 122.7 ng/L (< 51), respectively. Cervical ultrasound found a non-suspect multi-micronodular thyroid with no increase in parathyroid volume. 99mTc-MIBI parathyroid scintigraphy suggested right retrothyroid hyperfunctional parathyroid more or less in position P4 (Fig. 1).

Cervicotomy with right parathyroid exploration was associated to resection of the right P IV, which showed macroscopically increased volume. Histology found slight nodular hyperplasia.

Postoperative biological analysis found persistent PHPT: ionized calcium, 1.40 mmol/L; PTH, 93 pg/mL (< 38.8). Hormonal assessment for associated multiple endocrine neoplasia was normal.

Parathyroid scintigraphy was again performed, coupled to CT (99mTc-MIBI SPECT/CT), 2 months post-surgically. The sestamibi fixation facing the right fossa was unchanged; sestamibi fixation as seen at the left pyriform sinus was suggestive of a hyperfunctional ectopic parathyroid or of an adenopathy (Fig. 2).

Flexible endoscopy visualized a submucosal tumefaction of the left pyriform sinus (Fig. 3).

Direct suspension laryngoscopy was performed, with CO_2 laser lesion resection. Macroscopic examination of the specimen found a $1.8 \times 1 \times 1$-cm nodular lesion of tissular consistency. Anatomopathologic examination suggested adenoma, despite the absence of an identifiable residual ring of normal parathyroid tissue.

Postoperatively, blood values returned to normal: ionized calcium, 1.15 mmol/L; PTH, 10.1 ng/L; at 6 months' follow-up, there was no recurrence.

Discussion

PHPT involving ectopic pyriform sinus adenoma has been rarely reported, and often entails difficulties of diagnosis and treatment [4,5].

In the present case, initial scintigraphy was false-positive for right retrothyroid adenoma, showing a subtraction image of sestamibi fixation with right supralobular projection. Scintigraphy generally shows excellent specificity, with the false-positive risk basically limited to associated nodular thyroid pathology [6], as in the present case. Image fusion coupling single-photon emission CT to conventional CT scan (SPECT/CT) easily detects such false positives. It also greatly enhances diagnostic sensitivity to ectopic adenoma [7] and seems indispensable in patients having already undergone surgical exploration. It is indeed sometimes recommended as a systematic first-line examination [8].

Thus, retrospectively, on the second scintigraphic exploration, the right supralobular projection image could be interpreted as showing micronodular lesions with greater uptake of 99mTc-MIBI than of technetium alone. As the ultrasound and peroperative aspects of these micronodular lesions were in no way suspect, no surgical treatment was applied.

It is noteworthy that the first scintigraphic examination also failed to diagnose ectopic adenoma. The sensitivity of planar scintigraphy is around 87% to 94% [2,6]. Even so, on retrospective re-reading of the first results, slightly reinforced fixation projecting under and inward of the left submaxillary gland could be discerned on the planar images. This represented fixation of the ectopic adenoma, which, however, was overlooked due to the low degree of fixation, its unexpected location and the apparently more typical scintigraphic right P4 image.

Cervical ultrasound is indispensable: it also can reveal increased parathyroid volume and enables precise morphologic assessment of the thyroid gland. Thus, associated US-SPECT-CT performs better than the alternative imaging techniques, raising sensitivity to more than 90% and thus enabling even more precise selection of candidates for minimally invasive surgery [5,9].

Embryologically the P3 and P4 parathyroids are of endodermic origin, deriving respectively from the 3rd and 4th branchial clefts. Two types of ectopia have been reported: congenital, resulting from pathologic embryonic migration, and acquired, resulting from secondary migration under gravitational force. To these may be added embryonic defects in pharyngeal sac migration, a rare pathology in which vestigial parathyroid tissue may subsist in the pharynx. It is rare for adenoma to develop on such vestigial tissue; very few cases have previously been reported [10], and the present case is an addition to these.

Endoscopic CO_2 laser resection has, to the best of our knowledge, been described in only one previous report [4]. The technical characteristics of CO_2 laser enable precise non-hemorrhagic surgery and thus complete excision conserving the gland. This intra-cavity technique avoided further cervicotomy, thus minimizing surgery time and morbidity.