Case Report

Persistent renal hyperparathyroidism caused by intrathyroidal parathyroid glands

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Abstract

Renal hyperparathyroidism usually occurs in chronic renal failure patients on regular dialysis. However, renal hyperparathyroidism resulting from intrathyroidal parathyroid glands is an uncommon condition. We herein present the case of a 35-year-old woman who has been on hemodialysis for 20 years. She had renal hyperparathyroidism with generalized weakness and bone pain for 2 years. The patient initially underwent parathyroidectomy at a local institution, during which two large parathyroid glands were resected from the right side (no parathyroid glands were found on the left side); however, the surgical procedure was unsuccessful, and the patient had persistent renal hyperparathyroidism after the operation. She was then transferred to our hospital and ectopic intrathyroidal parathyroid glands were localized by neck ultrasonography and technetium-99m sestamibi scans with single-photon emission computed tomography imaging preoperatively. A left thyroid lobectomy was performed and two intrathyroidal parathyroid glands were found. The patient recovered uneventfully and her symptoms resolved. Therefore, clinicians should be aware of the possibility of renal hyperparathyroidism resulting from intrathyroidal parathyroid glands in cases where the renal hyperparathyroidism persists after parathyroidectomy.

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1. Introduction

Renal hyperparathyroidism occurs in patients with chronic renal failure. Approximately 15% of patients after 10 years and 38% of patients after 20 years of dialysis become refractory to medical treatment and therefore require parathyroidectomy.1,2 Persistent or recurrent renal hyperparathyroidism occurs in patients with ectopic or supernumerary parathyroid glands that are missed on initial parathyroidectomy. However, renal hyperparathyroidism resulting from the intrathyroidal parathyroid hyperplastic glands is uncommon.3 We herein report a rare case of a patient with persistent renal hyperparathyroidism resulting from intrathyroidal parathyroid glands. Good localization of intrathyroidal parathyroid glands is considered crucial preoperatively. Technetium-99 m (Tc-99m) sestamibi scans with single-photon emission computed tomography (SPECT) imaging and ultrasonography are useful techniques for accurate localization of intrathyroidal parathyroid glands.4 A thyroid lobectomy should be considered in situations where an intrathyroidal parathyroid gland is suspected to resolve persistent renal hyperparathyroidism.

2. Case report

A 35-year-old uremic woman has been on hemodialysis for 20 years. Her kidney transplantation failed when she was 28 years old. She had renal hyperparathyroidism with weakness,
fatigue, myalgia, bone pain, and pruritus for the past two years. The initial intact parathyroid hormone (iPTH) level was 930 pg/mL and her thyroid function test measurements were as follows: T3, 64.56 ng/dL (range, 86–187 ng/dL); thyroid-stimulating hormone, 10 IU/mL (range, 0.25–5.0 IU/mL); and free T4, 0.72 ng/dL (range, 0.8–2.0 ng/dL). She underwent a prior unsuccessful parathyroidectomy at a local institution, during which two large parathyroid glands were resected from the right side (no parathyroid glands were found on the left side). Two weeks after the initial operation, her iPTH level was 500 pg/mL and her symptoms persisted. Subsequently, her serum iPTH level continued to increase and was 3411 pg/mL with concurrently elevated serum calcium level (Ca\(^{2+}\), 12.1 mg/dL).

She was transferred to our hospital and neck sonography was performed, which showed a 1.5 cm × 1 cm × 1 cm hypoechoic nodule in the left upper pole of the thyroid and a 1 cm × 1 cm × 1 cm heterogeneous nodule in the left lower pole of the thyroid without other lesions consistent with parathyroid tumors (Fig. 1). The Tc-99m sestamibi scan with SPECT showed increased methoxyisobutylisonitrile (MIBI) uptake in the left thyroid bed in both early and delayed imaging phases, with a stronger uptake at the left upper pole of the thyroid (Fig. 2). Because of concordant MIBI and ultrasound findings, we suspected persistent renal hyperparathyroidism resulting from the left intrathyroidal parathyroid glands.

During the operation, we explored the left side of the neck, and found no parathyroid tumors. We then performed a left thyroid lobectomy based on the results of preoperative localization studies. We found two tumors in the left lobe of the thyroid (upper: 1.5 cm × 1.0 cm × 1.0 cm and lower: 1.0 cm × 1.0 cm × 0.8 cm). Frozen biopsy confirmed the excised mass to be parathyroid tissue (Fig. 3). Permanent pathological examination was performed, and the results showed intrathyroidal parathyroid glands with nodular hyperplasia. Postoperative serum iPTH level was 10.3 pg/mL and calcium was 9.2 mg/dL. Her symptoms resolved following the surgical procedure.

3. Discussion

Persistent and recurrent hyperparathyroidism are expected outcomes of subtotal parathyroid resection; however, its rate depends on the extent of initial resection and the duration of follow up. Remnant parathyroid tissue will grow, irrespective of whether it was left on purpose (as in a planned three-and-a-half gland subtotal parathyroidectomy) or could not be found during the initial operation (as in our patient). The reported rates of persistent and recurrent hyperparathyroidism are 10–12% for both subtotal parathyroidectomy and total parathyroidectomy with autotransplantation, and 0–4% for total parathyroidectomy without autotransplantation.\(^3\) Causes for

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**Fig. 1.** Two heterogeneous hypoechoic lesions in the (A) upper (1.53 cm × 1.0 cm, white arrow) and (B) lower (0.90 cm × 0.94 cm, white arrow) poles of the left lobe thyroid gland.

**Fig. 2.** Increased methoxyisobutylisonitrile (MIBI) uptake in the left thyroid bed.
The intrathyroidal parathyroid glands are usually found incidentally after thyroidectomy with a reported rate of 1.4–3.2%. There is a continuing debate about the embryologic origin of intrathyroid parathyroid glands. Most believe that they are embryologically inferior parathyroid glands. In a previous study of 43 patients with intrathyroidal parathyroid glands, Proye et al reported three superior and 44 inferior glands. Our patient with simultaneously superior and inferior intrathyroidal parathyroid glands is distinctively unusual. Other more common locations for ectopic superior parathyroid glands are the retroesophageal–retropharyngeal area and the posterior mediastinum.

Ultrasoundography is a convenient, noninvasive, and inexpensive tool for preoperative localization. Although thyroid nodules in patients with renal hyperparathyroidism are very common, a thyroid nodule detected by ultrasonography during an examination for patients with parathyroid disease could be an intrathyroidal parathyroid gland. When in doubt, fine-needle aspiration biopsy (FNAB) with cytology and iPTH assay should be performed, which can easily distinguish a thyroid nodule from an intrathyroidal parathyroid. Ultrasoundography is a very useful preoperative localization study for hyperparathyroidism; it is also the most useful for the study of intrathyroidal parathyroid glands. However, it is indeed difficult to detect intrathyroidal parathyroid glands when the thyroid nodule is not big enough to be eligible for performing further studies (e.g., sono-guided FNAB) based on the criteria suggested by the American Thyroid Association Guidelines and National Comprehensive Cancer Network Guidelines.

The Tc-99m sestamibi scan is also a useful modality for preoperative localization. It is, however, less sensitive for renal hyperparathyroidism (76.2% for nodular type and 28.6% for diffuse type) than for primary hyperparathyroidism (88.4% for solitary adenoma and 44.46% for multiple adenoma). The sensitivity of the scan results can be increased by combining it with ultrasonography if the two studies are concordant. Other studies such as computed tomography/magnetic resonance imaging or selective venous sampling can also be used in patients with persistent/recurrent renal hyperparathyroidism. However, thyroid lobectomy should be considered in situations where an intrathyroidal parathyroid gland is suspected perioperatively. Before ultrasound became a routine preoperative examination, a “blind thyroid lobectomy” was usually performed when the surgeon is exhausted in the search for a missing parathyroid gland on the same side (after doing a cervical thymectomy, explored the tracheoesophageal groove, and opened the carotid sheath). At present, with ultrasound being a routine preoperative examination, the surgeon may consider performing a thyroid lobectomy earlier when a missing parathyroid gland is on the same side in which there is a hypechoic “thyroid nodule”, which has not had been confirmed to be a colloid nodule by a prior FNAB. An enucleation of the intrathyroidal parathyroid, as an enucleation of a thyroid nodule, is a poor treatment with potential problems of rupture and complications associated with possible need for reoperation.

Our patient had uncertain ultrasonography examination over the usual location and ectopic sites of parathyroid glands. Although Tc-99m sestamibi scans can help to confirm the ultrasound findings, occasional follicular thyroid tumors can cause false positives. If an ultrasound-guided FNAB is considered, the aspirate should be sent for both cytological analysis and iPTH assay. In general, measuring iPTH in the aspirate is more accurate than performing a cytological examination. However, one should be aware of the possibility of needle-track seeding causing iatrogenic parathyroidosis.

Intrathyroidal parathyroid glands can be missed during the initial operation, which could subsequently result in persistent/recurrent diseases. This rare anatomic anomaly should be considered when a parathyroid gland is not identified but a thyroid nodule was present on preoperative ultrasound. For reoperations, preoperative localization studies including ultrasonography and Tc-99m sestamibi scans are crucial. Thyroid lobectomy should be considered during the initial operation or re-operation when intrathyroidal parathyroid is suspected.

References


