

Rare concurrence of ectopic intrathyroidal parathyroid gland and papillary thyroid carcinoma within a thyroid lobe

A care-compliant case report

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Abstract

Rationale: The inferior parathyroid glands derive from the third branchial pouch and seldom ectopically migrate to thyroid gland, mediastinum, thymus. Ectopic intrathyroidal parathyroid gland (ETPG) is rare and it cannot be consistently detected by any imaging examinations. The unusual ETPG is easily resected during thyroidectomy, which might cause postoperative hypoparathyroidism. Auto-transplantation of even a parathyroid gland can restore parathyroid function to some extent. Until now, there are not any guidelines on the management of ETPG.

Patient concerns: Without any symptoms, a 27-year-old female patient was admitted to our hospital for a thyroid nodule. Thyroid ultrasonography demonstrated a hypoechoic nodule, measuring 0.6 cm × 0.6 cm × 0.7 cm, with multiple punctate microcalcifications within the right thyroid lobe. Thyroid function tests were normal. The invasive fine needle aspiration cytology for the thyroid nodule was rejected by patient.

Diagnoses: The patient was firstly diagnosed as right thyroid hypoechoic nodule, which was pathologically proved to be papillary thyroid carcinoma (PTC) finally.

Interventions: The patient underwent right thyroidectomy, central compartment neck dissection, parathyroid exploration, and auto-transplantation of the normal ETPG.

Outcomes: The right thyroid hypoechoic nodule was diagnosed as PTC by pathological examination. The inferior parathyroid gland could not found in normal locations. On the cut surface of excised thyroid lobe, a yellow and soft nodule was discovered and it was pathologically diagnosed as a normal parathyroid gland. In the end, auto-transplantation of the normal ETPG was conducted. Postoperative follow-ups found both serum calcium and parathyroid hormone tests were normal.

Lessons: ETPG in a patient with PTC is easily overlooked and unexpectedly excised for its rarity. ETPG brings about a failed parathyroid exploration. However, within the thyroid parenchyma, it might be occasionally discovered by cutting a thyroid lobe into slices. Auto-transplantation of normal ETPG is necessary.

Abbreviations: ETPG = ectopic intrathyroidal parathyroid gland, PTC = papillary thyroid carcinoma.

Keywords: auto-transplantation, ectopic intrathyroidal parathyroid gland, hypoparathyroidism, papillary thyroid carcinoma

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1. Introduction

During embryogenesis, the inferior parathyroid glands may migrate to variable anatomic locations such as superior mediastinum, carotid sheath, thymus, and even thyroid.^[1,2] The incidence of ETPG within the thyroid parenchyma was reported to be 0.2% from a large anatomical series study.^[3] Compared with ETPG, ectopic intrathyroidal parathyroid lesions, such as adenoma or carcinoma, can be more easily diagnosed through classical symptoms of hyperparathyroidism, imaging examinations, or fine needle aspiration cytology.^[4,5] Since late ischemia of remaining parathyroid glands probably happens, routine auto-transplantation of unintentionally excised parathyroid glands during thyroidectomy can restore parathyroid function and reduce the risk of postoperative hypoparathyroidism.^[6] We report the ETPG in a young patient with papillary thyroid carcinoma (PTC) and introduce some guidelines on the management of ETPG during thyroidectomy.



Figure 1. Thyroid ultrasonography demonstrated a hypoechoic nodule with multiple punctate micro-calcifications within right thyroid lobe (arrow).

2. Case report

The institutional review board of Peking Union Medical College Hospital approved this work. The patient has provided informed consent for publication of the case. The 27-year-old female patient, an office clerk, was admitted to the department of general surgery for a right thyroid nodule. Thyroid ultrasonography shown only a hypoechoic nodule, measuring 0.6 cm × 0.6 cm × 0.7 cm, with multiple punctate micro-calcifications within right thyroid lobe (Fig. 1, arrow), and there were no enlarged lymph nodes in the bilateral neck areas. Symptoms, physical examination, and past medical history were negative and family history was insignificant for similar sickness. Thyroid function tests were normal. Serum total calcium level was 2.63 mmol/L (normal range 2.13–2.70 mmol/L). She rejected examination of fine needle aspiration cytology. The diagnosis was initially considered as right thyroid hypoechoic nodule: the suspicious PTC. She underwent right thyroidectomy and central compartment neck dissection. Then, the hypoechoic nodule within the excised thyroid lobe was separately sent to intraoperative pathological examination. The hypoechoic nodule was pathologically diag-

nosed as PTC in the end. Routine parathyroid exploration failed to discover inferior parathyroid gland in normal locations. The excised lymphoid tissue of central compartment and surface capsule of thyroid lobe were also explored, but no suspicious parathyroid tissue was discovered. Then, we horizontally cut thyroid parenchyma into slices and found a well demarcated, yellow, and soft nodule inside, measuring about 0.5 cm × 0.9 cm (Fig. 2A, rectangular box). Small part of the yellow nodule (about 0.1 cm × 0.2 cm) (Fig. 2A, “a”) was cut off and it was proved to be normal parathyroid gland tissue by pathological examination (Fig. 2B). Finally, the remaining normal parathyroid gland tissue (Fig. 2A, “b”) was rapidly transplanted into ipsilateral sternocleidomastoid muscle. Postoperative serum total calcium level was 2.16 mmol/L (normal range 2.13 ± 2.70 mmol/L) and parathyroid hormone level was 16.2 pg/mL (normal range 12.0–68.0 pg/mL) 1 day after surgery. Both 1 month and 3 months after surgery, serum total calcium and parathyroid hormone level were in normal range.

3. Discussion

We report an unusual thyroid lobe with coexistent ETPG and PTC in a young patient. In our case, ETPG could not be detected by thyroid ultrasonography. It was occasionally found by exploring thyroid parenchyma, and the diagnosis of ETPG was established by pathological examination. To the best of our knowledge, it is not rare that parathyroid glands are embedded on surface capsule of thyroid gland. However, they seldom arise in ectopically located thyroid parenchyma. ETPG may deteriorate into ectopic intrathyroidal parathyroid adenoma or carcinoma,^[4,5] but the deterioration mechanisms are not clear so far. Compared with above ectopic intrathyroidal parathyroid lesions, ETPG generally cannot bring about hyperparathyroidism and it cannot be consistently detected by imaging examinations such as ultrasonography, computed tomography, or radionuclide imaging.

Ultrasonography is often applied to diagnosis of parathyroid lesions such as adenoma or carcinoma. However, there are quite little studies^[7,8] upon ultrasonic characteristics of normal parathyroid glands: near the lower pole and outer edge of thyroid lobe, normal parathyroid glands are often located

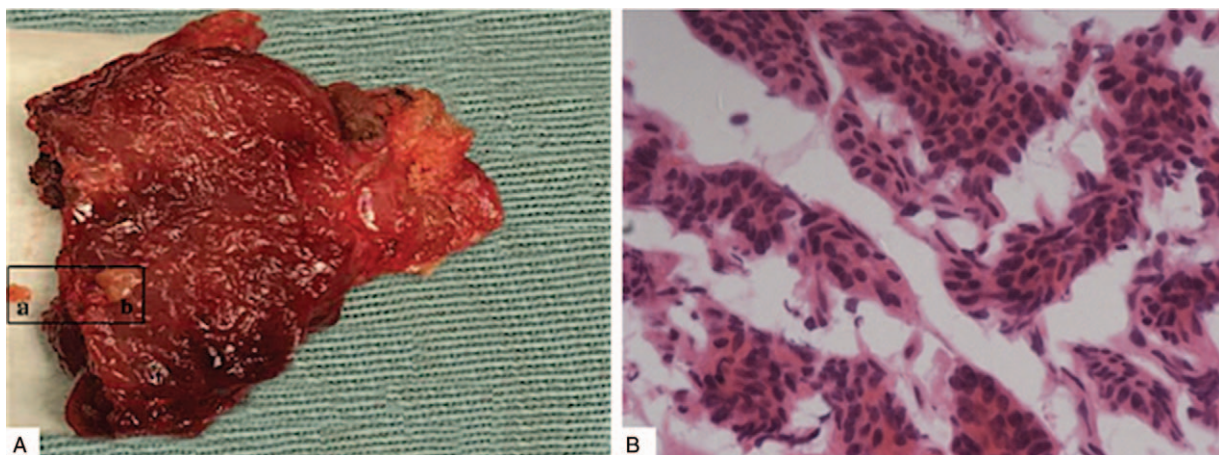


Figure 2. A well demarcated, yellow, and soft nodule was discovered within the excised right thyroid lobe (A, rectangular box); small part of soft nodule (A, “a”) was cut off and it was proved to be normal parathyroid gland tissue by intraoperative pathological examination (B). The remaining tissue (A, “b”) was transplanted into ipsilateral sternocleidomastoid muscle.

between trachea and common carotid artery. Most of them have an oval shape, clear boundary, and uniformly strong echo. The Color Doppler flow imaging showed no or less blood flow, and ultrasound elastography showed that normal parathyroid glands are slightly softer than thyroid glands. The ultrasonic display rate of normal parathyroid glands varies from literature to literature, which is even as low as 8.3%.^[9] However, it is very challenging and difficult to detect normal ETPG due to several factors, including its rarity, small volume, tissue interference of thyroid parenchyma, and lack of clinical experience.

So far, normal ETPG is exceedingly rare with about four cases^[10–13] previously reported, but concurrent ETPG and PTC inside a thyroid lobe is the 1st in China. Velimezis et al^[10] reported a thyroid lobe with a concurrence of PTC, ETPG, and ectopic intrathyroidal thymus tissue. Kim et al^[11] reported a patient with a history of PTC in the right thyroid lobe, but the patient also had an unusual association of ETPG and ectopic intrathyroidal thymus tissue in the left thyroid lobe. Bernd et al^[12] reported a concurrence of ETPG and papillary microcarcinoma, and Abdou et al^[13] reported a close association of ETPG and thyroid amyloidosis tissue inside a thyroid lobe. In our case, we introduce some guidelines on management of normal ETPG during thyroidectomy. The occurrence of ETPG is one of main factors for failed parathyroid exploration. If parathyroid glands fail to be detected in the normal positions, the resected thyroid lobe should be the target of exploration. It is suggested that the thyroid parenchyma should be horizontally cut into slices. When a yellow, oval, soft, and well demarcated nodule is discovered, small part of this yellow nodule should be cut off and be sent to intraoperative frozen pathological examination. The rest should be soaked into normal saline for preservation. Once it is histologically proved to be normal parathyroid tissue, the rest should be disposed prudently. However, the ETPGs, which were reported by above 4 cases,^[10–13] were not discovered by exploring thyroid parenchyma during thyroidectomy. Therefore, these ETPGs have been thrown away unexpectedly. To the best of our knowledge, auto-transplantation of normal ETPG in the patient with PTC has not been reported so far. Considering late ischemia of remaining parathyroid glands, auto-transplantation of even a parathyroid gland can offer insurance for preserving parathyroid function.^[6] In our case, the normal ETPG was discovered during thyroidectomy and it was immediately transplanted.

Some profound lessons are in our report. ETPG is easily overlooked for its rarity, and it cannot be sensitively detected by parathyroid ultrasonography. ETPG will cause a failed

parathyroid exploration. If surgeons accustom themselves to exploring the excised thyroid lobe carefully, the discovery rate of ETPG will increase. Auto-transplantation of normal ETPG may prevent postoperative hypoparathyroidism and hypocalcemia.

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