Unusual Localizations of the Ectopic Parathyroid Gland on the Anterior Aspect of the Thyroid: A Report of Two Cases.

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Abnormally located or supernumerary parathyroid glands are not rare. Ectopic parathyroid glands can be seen anywhere between the mandibular angle and the mediastinum. In this report two cases with ectopic parathyroid glands are presented. First case is a patient presenting with persistent hyperparatyhroidism who had an ectopic fifth parathyroid on the median line, superior to thyroid's isthmus in front of the cricoid cartilage displaying normal histological findings and a sixth gland embedded in the pericardiac tissue which is an adenoma causing the hyperparathyroidic state. Second case is a patient operated for toxic multinodular goiter with an ectopic parathyroid gland on the anterior aspect of the thyroid isthmus' superior border with a vascular pedicule arising from the mediastinum. In this patient, the gland anterior to the isthmus is thought to be the ectopically located left lower parathyroid which was not detected in its usual localization.

Key words: **Ectopic parathyroid, supernumerary parathyroid, thyroid**

Introduction

Parathyroid (PT) glands generally lie close to the posterior surface of thyroid lobes and there are usually four PT glands in humans (1). Abnormally located or supernumerary PT glands are not rare. Their variable locations in relation to the thyroid gland and other structures of the neck should be well known. Because undetected supernumerary or ectopic PT glands have a high risk of persistent and recurrent hyperparathyroidism, these regions must be carefully inspected during neck exploration (2).

In this report two cases are presented. The first case is a patient with ectopic and supernumerary PT glands. The fifth PT was normal and ectopic, sixth one was an ectopic adenoma. The second case is a patient with an ectopic fourth PT. All these anomalies have not been reported to our knowledge before in the literature.

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Case 1

A 50 year-old-female patient was hospitalized for incisional hernia repair. Medical history revealed well controlled hypertension for a duration of 6 months. All routine preoperative biochemical analysis were normal except for serum calcium (Ca) and phosphorus (P) levels. In our clinic serum Ca and P levels are routinely checked in hypertensive patients. Serum Ca and P levels were 12.6 mg/dl (8.08-10.4), and 2.7 mg/dl (2.7-4.5) respectively. After the detection of the high abnormal control values, PTH and 24 hour urinary calcium were measured. PTH was 187 pg/ml (9- 80) and urinary calcium was 300 mg/day (100-300). Ultrasound scan of the neck revealed only hypoechogenic

nodules of 8.9mm and 9.0mm at the right lobe's anterior partand 4.7 mm and 8.0mm at the left lobe's posterior part. Tc99-MIBI scan did not show any abnormal activity of the PT glands. With these findings, the patient was diagnosed with primary hyperparathyroidism (PHPT) and bilateral cervical exploration was performed. Four PTs of normal size were detected in the usual localizations. Samples for frozen section were taken with applying clips from right upper and lower PTs. After checking the blood supply of the remnants the same procedure was performed on the contralateral glands. Frozen section confirmed that all the samples were PT tissue. Other possible sites for ectopic PT glands were inspected and servical thymectomy was performed. In the inspected area and thymectomy specimen there was no macroscopic tissue resembling PT. Regarding the presence of palpable hard thyroid nodules and the probability of intrathyroidal PTs, it was decided to perform thyroidectomy. During the mobilization of the isthmus, a yellow brown, oval nodule (5x3x2 mm in diameter) was found embedded in the soft tissue superior to thyroid isthmus,in front of the cricoid cartilage in the midline (Fig.1) Frozen section examination showed that it was also PT tissue. After the completion of near total thyroidectomy operation was terminated. Permanent pathological examination showed that the four PT samples from the normal loci and the fifth gland which was excised from an ectopic localization had normal histologic features. In the postoperative period biochemical analysis showed that the patient had persistent PHPT. A single photon emission computed tomography (SPECT) was indicated due to persistent PHPT which revealed a mass consistent with parathyroid adenoma 3-4 cm beneath the anterior wall of the thorax, 3 cm to the right of the midline in the anterior mediastinum. Median sternotomy was performed and gamma probe guided exploration was utilized to detect and excise a lesion which was over 1 cm in diameter that was embedded in the pericardiac fatty tissue. Frozen section confirmed that the excised mass was PT tissue. In the early postoperative period serum Ca, P, and PTH levels returned to normal and ten months after the operation the patient was in a normocalcemic state.

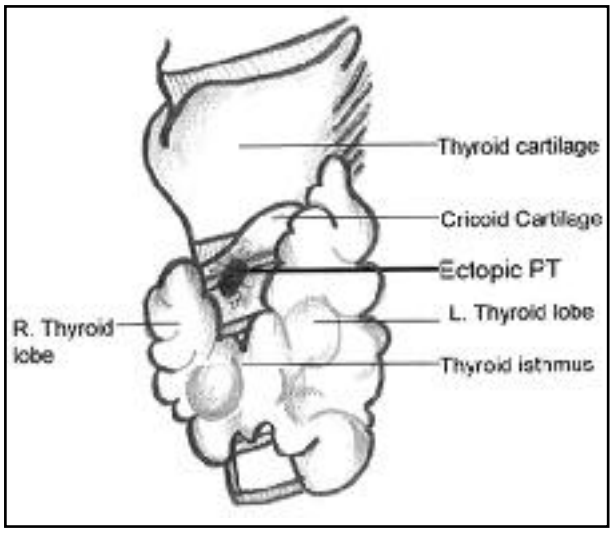

Figure 1. Illustration of the localization of the ectopic fifth PT gland in the soft tissue on the median line, superior to thyroid's isthmus in front of the cricoid cartilage.

Case 2

An operation was planned for a 60 year old female patient with toxic multinodular goiter (MNG) in euthyroid state with the findings of a 2.5 cm dominant nodule in the left thyroid lobe and bilateral multiple nodules detected by physical examination, USG and scintigraphy. In the operation, multiple nodules were observed on both thyroid lobes. An oval, light brown mass with a diameter of 5 mm was detected on the anterior of the thyroid isthmus' superior border. The mass had one single vascular pedicule arising from the mediastinum and running over the isthmus (Fig. 2). A 1 mm biopsy was taken from the opposite side vascular entrance for frozen section, which was identified as the PT tissue. The ectopic PT gland was dissected with its vascular pedicle. Bilateral upper and right lower

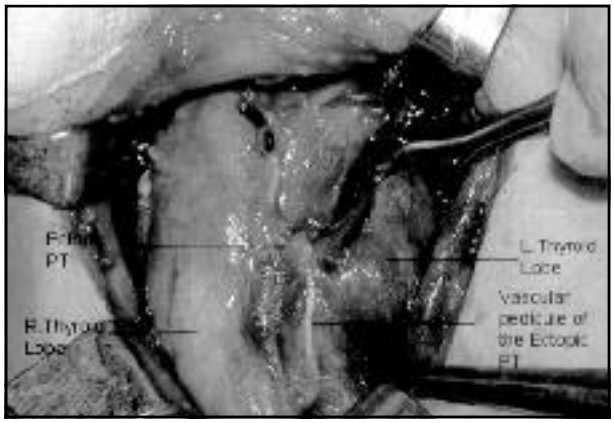

Figure 2. Ectopic PT gland of case 2 on the anterior of the thyroid isthmus' superior border.

parathyroid glands were detected in the normal position and were observed to have arterial supplies provided by the branches of the inferior thyroid artery. Left lower parathyroid gland was not found in the usual localization. Following left total and right subtotal thyroidectomy, the ectopic PT with its intact vascular pedicle was placed on the lateral left side of recurrent laryngeal nerve. Hypocalcemia was not detected in the postoperative period.

Discussion

Ectopic parathyroid glands can be seen anywhere between the mandibular angle and the mediastinum (3). Akerström (1) and Wang (4) reported that 1% of superior parathyroid glands and 2% of inferior glands were observed in ectopic locations in autopsy studies. On the other hand, in a prospective study on hyperparathyroidic patients by Thompson et al (3), 12% of the adenomas of the right and, 16% of adenomas of the left inferior PT were found in an ectopic location, such as thymus, carotid sheath, or lower pole of the thyroid. In one third of the adenomas of the superior PTs were found in abnormal locations that is in retropharyngeal, retroesophageal, paraesophageal, tracheoesophageal grooves and the tracheal bifurcation. Ectopic localization of PT in the interthyrotracheal groove, on the strap muscle, within nervus vagus, in the left posterior of the neck and in the sinus pyriformis have also been reported in patients with primary hyperparathyroidism (2,5-9).

On the other hand in the autopsy series of Akerström et al (1), 13 percent of the patients had a supernumerary gland. In approximately two thirds of the patients, the supernumerary gland was found below the thyroid in association with the thymic ligament or the thymus on either side. In approximately one third of the patients, the supernumerary gland was found in the vicinity of the thyroid between the two other glands. In addition, the inferior glands located in the thymus and further down in the mediastinum were seen in 2 per cent of patients, and only 0.2 percent of patients were encountered below the thymus in the anterior mediastinum.

In the first case four glands were found in the normal localizations and histologically all were normal PT tissue. The fifth one which was superior to thyroid isthmus and in front of the cricoid cartilage also displayed normal histologic appearance (Fig. 1). In this case PHPT was due to the adenoma of the sixth PT gland embedded in the pericardiac tissue which was supplied by the multiple thin artery arising from the fatty tissue. Because blood supply of ectopic mediastinal gland usually comes from inferior thyroid artery. Very rarely, mediastinal glands may be supplied by the internal mammarian artery (11). In our knowledge ectopic localizations of the fifth PT in the first case presented also has not been reported in the literature previously. Unusual arterial supply of the sixth PT which was a mediastinal gland was also not reported up to date.

Second case in this report as mentioned above had MNG with an ectopic PT on the anterior of the isthmus and other three glands in their normal localizations. Thus, this finding may indicate that the fourth gland is an ectopic gland. Because the patient had undergone thyroidectomy for MNG, thymus and the other probable ectopic PT locations were not explored. This localisation of PT has not been reported to our knowledge in previous autopsy or HPT series. Another extraordinary aspect of the second case was the blood supply of the gland, which was provided by a stalk arising from mediastinum (Fig. 2). The vascular supply to the parathyroid glands is usually from the inferior thyroid artery, but it can arise from the superior thyroid artey, the lowest thyroid artery (thyroidea ima), and arteries in the larynx, trachea, esophagus, or mediastinum or from anastomoses between the vessels (10). This anomaly (ectoic location servical PT) was also not reported up to date to our knowledge.

Ectopic parathyroid glands are a major cause of persistent and recurrent hyperparathyroidism (2). Parathyroid glands must be identified during cervical exploration and the absence of a gland in one of the usual locations should indicate ectopic localization and the exploration should be carried on regarding this possibility. When four parathyroid glands are detected in the usual locations in a patient with hyperparathyroidism, possibility of a supernumerary gland should be kept in mind. In addition to the exploration of common ectopic localizations (12), we believed that exploration around thyroid isthmus should also be performed.

Identifying ectopic PT glands located close to the thyroid gland is important in the thyroid operations

as well. PT glands located around the thyroid may be confused with fat lobules, accessory thyroid nodules or lymph nodes. Physical characteristics of the parathyroid like the color, shape, consistency and the vascular supply of the gland may help to differentiate between these tissues (12,13). When a round, lobulated mass is encountered around the isthmus during thyroidectomy as in case 2, possibilty of an ectopic PT gland should be kept in mind and identification of tissue with frozen section should be done before resection. If frozen section shows that it is PT tissue, every effort should be done in order to preserve it with its intact blood supply. Otherwise, the tissue in 1 mm slices should be inoculated to the sternocleidomastoid or forearm's muscles after resection.

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