Nonadenomatous nonencapsulated thymic parathyroid tissue concomitant with primary hyperparathyroidism due to ectopic parathyroid adenoma

SUMMARY
Primary hyperparathyroidism due to ectopic parathyroid adenoma is not infrequent. Primary hyperparathyroidism caused by unusual thymic nonadenomatous nonencapsulated parathyroid tissue has been reported before. Both can cause unsuccessful neck explorations. Here we presented for the first time a patient with hyperparathyroidism due to ectopic parathyroid adenoma concomitant to the presence of thymic nonadenomatous nonencapsulated parathyroid tissue.

INTRODUCTION
Minimally invasive parathyroidectomy is generally used for the treatment of primary hyperparathyroidism in patients with parathyroid adenoma (1). If there is doubt in localization, open bilateral neck exploration is considered the standard option (2). Ectopic parathyroid adenoma is a common cause of failed parathyroidectomies (3). Incidence of ectopic parathyroid glands is up to 10%, and the majority of them are inferior parathyroid glands because of abnormal migration during embryogenesis (4).

Embryologically, the thymus migrates caudally escorted by the inferior parathyroid glands. Parathyroid tissue may descend up to thymus. Primary hyperparathyroidism due to nonadenomatous unencapsulated parathyroid tissue has been previously reported (5). Here, we present, for the first time, an extremely rare condition of primary hyperparathyroidism. To our knowledge, this is the first case regarding the coexistence of thymic nonadenomatous nonencapsulated parathyroid tissue and an ectopic parathyroid adenoma in the tracheoesophageal groove.

CASE REPORT
A 20-year-old female patient presented long-term bone pain and fatigue. The patient’s serum calcium was 12.5 mg/dL (normal range: 8.6-10.3 mg/dL), phospho-
Nonadenomatous thymic parathyroid tissue concomitant with ectopic parathyroid adenoma

The majority of patients (80%-85%) with primary hyperparathyroidism have a solitary adenoma (6). In 80%-85% of these patients, the adenomas are located in typical anatomical localizations, and in 15%-20% of the cases they are ectopic (7). Superior parathyroid glands along with the thyroid glands lateral lobes develop from the 4th branchial pouch, whereas inferior glands, along with the thymus, develop from the 3rd branchial pouch and move caudally to their normal anatomical location (8). During this caudal migration, any abnormality may result in the ectopic placement of these glands. There are two types of ectopic localization. The first type is due to mechanical factors, and occur as a result of large adenomas. This is usually observed in the superior glands. In the second type, the ectopic location is due to abnormal organogenesis. This second type is usually observed in the inferior glands (9). Superior ectopic glands may be located in the retropharyngeal, retroesophageal and tracheoesophageal regions. Inferior ectopic glands may be retroesophageal, intrathyroidal, intrathymic regions, within the carotid sheath, thyrothymic ligament, within the aortopulmonary window in the mediastinum, and within the pericardium (9).

In our case, the patient had a solitary adenoma located ectopically within the tracheoesophageal sulcus on the right side, and thymic nonencapsulated parathyroid tissue on the left side.

The lesion, which was observed on the left in the scintigraphy, disappeared after the first operation, and serum parathyroid hormone and calcium levels were slightly lower after first operation. Furthermore, the ectopic adenoma suppressed all other normal glands but did not appear to suppress the thymic parathyroid tissue. Therefore, thymic nonadenomatous parathyroid tissue may have contributed to the hyperparathyroid condition in this case. Another case report described nonadenomatous unencapsulated parathyroid tissue as a cause of persistent primary hyperparathyroidism (5). The association of the ectopic parathyroid glands and the thymus may be due to their common embryologic origin from the pharyngeal pouch or common genetic abnormality. However, we cannot evaluate if the nonadenomatous thymic parathyroid tissue would be able to maintain the hypersecretion state if the ectopic adenoma was resected first.

Gamma probe can help surgeons to locate adenomas (10,11). In the second operation of the case that we presented, we administered a low dose radiotracer, used the intraoperative gamma probe, successfully located the ectopic adenoma, and removed it without the need of frozen section to confirm it.

There may be more than one ectopic parathyroid lesion located in different places, as occurred in this case. Also, it must be kept in mind that there may be ectopic hypercellular parathyroid lesions in thymus rather than distinct adenomas. Careful histological examination is of paramount importance in resected thymus tissue.
Figure 1. (A) Preoperative $^{99m}$Tc-sestamibi dual phase parathyroid scintigraphy. Early (a) and delayed (b) phase images reveal two foci of persisted increased activity at the inferior tip of both lobes of the thyroid gland; however, pathological activity focus on the left is located more to the bottom than to the right. (B) Control $^{99m}$Tc-sestamibi dual phase parathyroid scintigraphy, 3 months after the first operation. Early (a) and delayed (b) phase images show a persistent focal uptake at the inferior part only in the right thyroid lobe.

Figure 2. Microscopic view in the histopathological examination of thymus resection material. Nonadenomatous cellular parathyroid tissue in the thymus (H&E, obj x 25) (Small, white arrow points to nonadenomatous cellular parathyroid tissue. Big, black arrow points to thyMIC tissue).
Disclosure: no potential conflict of interest relevant to this article was reported.

REFERENCES


Nonadenomatous thymic parathyroid tissue concomitant with ectopic parathyroid adenoma