
CASE REPORT

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DUAL THYROID ECTOPIA WITH A NORMALLY LOCATED PRETRACHEAL THYROID GLAND: CASE REPORT AND LITERATURE REVIEW

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Abstract: *Background.* Only 1 case with dual thyroid ectopy and a normally located thyroid gland has been reported.

Methods. We present the case of a 71-year-old woman who had 1-sided lateral neck swelling in the right submandibular space, which grew and doubled in size within 1 year. A CT scan demonstrated 2 right submandibular, lobulated, heterogeneously enhanced masses, an ectopic lingual thyroid, and bilateral thyroid goiter. A Tc-99m sodium pertechnetate thyroid scan revealed a cold area in the right lateral neck region below the submandibular gland, an ectopic lingual thyroid, and bilateral pretracheal thyroids. She underwent total thyroidectomy and en bloc excision of right neck masses. Thyroxin was prescribed following the surgery.

Results. Pathological diagnoses were ectopic thyroid tissue with goitrous change and bilateral thyroid goiter.

Conclusion. This report demonstrates that dual ectopic thyroid tissue accompanying a normally located thyroid gland can exist and should be differentiated from head and neck malignancies. ©2007 Wiley Periodicals, Inc. *Head Neck* 29: 885–888, 2007

Keywords: ectopic thyroid; dual thyroid ectopy; lateral neck masses; submandibular mass; neck swelling

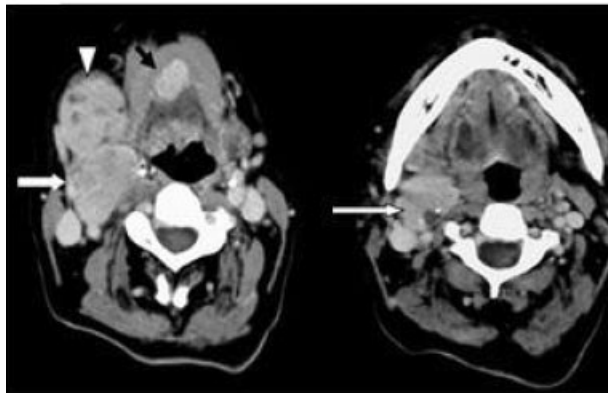
The thyroid gland is the first endocrine gland that develops in the human embryo. A mature thyroid gland has 2 hormone-producing cells, thyroid follicular cells, and parafollicular cells, also called C cells. Thyroid follicular cells are derived from the thyroid anlage, whereas parafollicular cells arise from ultimobranchial bodies. The thyroid anlage, which originates from the floor of the primitive pharynx at the end of embryonal week 3, is of endodermal origin and is located on the midline of the embryonic mouth cavity in its posterior section. However, ultimobranchial bodies, which form in embryonal week 4 from the fourth pharyngeal pouch, are of neuroectodermal origin and are located symmetrically on the lateral sides of a developing neck. The thyroid anlage is connected to the primitive pharynx by the thyro-

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A.



B.

C.

FIGURE 1. Contrast-enhanced axial CT scan. (A) Normally located bilateral goiters. (B) Right submandibular lobulated heterogeneously enhanced masses with hypodense foci (white arrow and arrowhead). Mouth floor lingual thyroid (black arrow). (C) The posterosuperior mass had spotty calcifications and extended to the right parapharyngeal space (white arrow).

glossal duct while descending. The duct starts to break down in embryonal week 5. During migration, the thyroid anlage develops into a bilobed structure. At embryonal week 7, the thyroid anlage finally descends to the level of second and third tracheal cartilages and fuses with the lateral ultimobranchial bodies.^{1,2} The thyroid primordium (or a portion of it) occasionally fails to descend via the normal pathway and results in an ectopic thyroid. Ectopic thyroid tissue is a rare entity, likely resulting from aberrant migration or developmental defects. Ectopic thyroid tissue can be found at any location along the migration path from the foramen cecum to the mediastinum with or without a coexisting normally located thyroid gland.³ Dual thyroid ectopy occurs rarely; only 1 dual ectopic thyroid case with a normally located thyroid gland has been reported.⁴

CASE REPORT

A 71-year-old woman was seen with a right upper lateral neck mass in the submandibular area present for roughly 1 year. At the age of 50, the

patient had undergone surgery for nodular goiter in another hospital, and some of thyroid tissue was excised. She was diagnosed with a benign thyroid lesion at that time, but the detailed information about the surgery was not available. About 1 year previously, she visited the metabolism outpatient department at Chang Gung Memorial Hospital because of the right submandibular mass. She denied any trauma history. Physical examination identified an elastic, mobile, nontender mass, 4 cm × 4 cm in size. Thyroid ultrasound examination and fine-needle aspiration cytology of the mass demonstrated adenomatous goiter. In addition, the thyroid hormone profiles were normal, including thyrotropin and free T₄. Thereafter, she received regular follow-up at the cardiovascular outpatient department for hypertension and was not treated for the thyroid lesion.

However, the right neck mass began to grow progressively. She reported occasional pain and moderate dysphagia. Ultrasonographic examination indicated bilateral thyroid goiter and right lateral neck mass. The mass was 8 cm × 6 cm and heterogeneous with moderate echogenicity. The lesion was not connected with the right thyroid tissue. Aspiration cytology revealed some degenerated follicular cells, follicular cells with focal atypia, and a few cell groups with solid



FIGURE 2. Contrast-enhanced coronal CT scan demonstrated a right lateral neck lobulated heterogeneously enhanced mass with hypodense foci (black arrow). Bilateral thyroid glands (white arrows). One clear margin between the mass and the right thyroid gland.

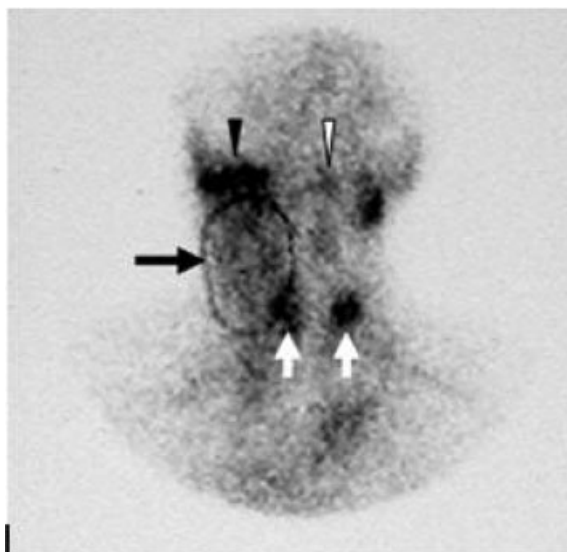


FIGURE 3. A Tc-99m pertechnetate thyroid scan. A cold area existed at the right lateral neck within the circle (black arrow). Mouth floor lingual thyroid (white arrowhead). Bilateral normally located thyroid glands (white arrows). Right submandibular gland was displaced upward (black arrowhead).

sheets mixed with possible colloid-containing follicles. Laryngoscopic examination demonstrated a normal upper aerodigestive tract. Head and neck CT scan with contrast-enhanced images disclosed 1 anteroinferior and 1 posterosuperior lobulated heterogeneously enhanced mass with hypodense foci at the right upper neck. The lesion was extended upward to the parapharyngeal space and downward to the lateral neck area near the upper pole of the right thyroid gland. Spotty calcification was also observed. Additionally, 1 lingual thyroid and enlargement of the bilateral thyroid glands with hypodense foci were found (Figures 1 and 2). The Tc-99m sodium pertechnetate thyroid scan demonstrated 1 slightly inhomogeneous ectopic thyroid tissue in the mouth floor, bilateral functioning thyroid glands, and a cold area in the right lateral neck area displacing the right submandibular gland upward (Figure 3).

The patient underwent surgery under the impression of the ectopic thyroid lesion; however, malignancy with cervical lymph node metastasis could not be completely excluded. Intraoperative findings demonstrated that both masses had goitrous change without invasion to adjacent tissues and were not associated with the right thyroid tissue. The bilateral thyroid glands were also enlarged with goitrous change. The upper pole of right thyroid and superior thyroid vessels were intact. The patient underwent total thyroidec-

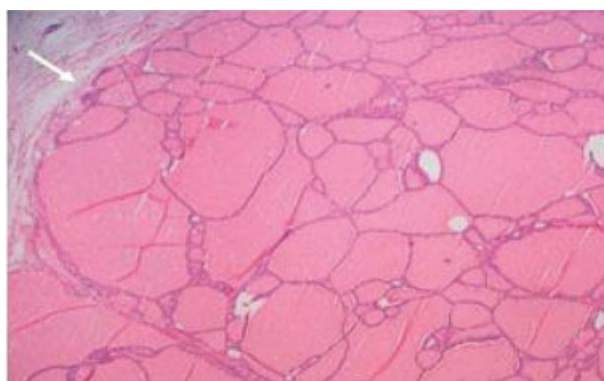
tomy and en bloc excision of the right submandibular lesion. The patient recovered uneventfully after the operation and underwent regular follow-up with prescribed thyroxin replacement.

PATHOLOGIC DIAGNOSIS

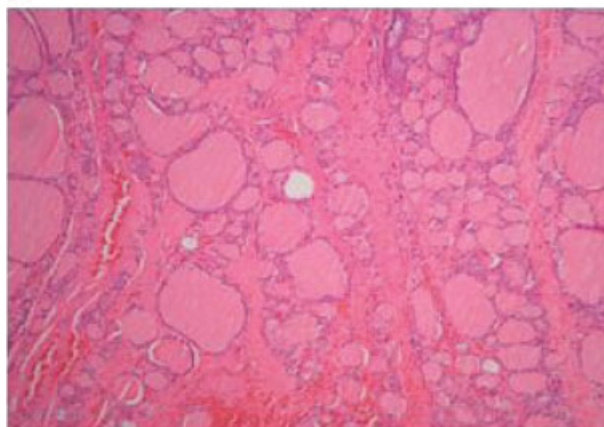
The right lateral neck masses and bilateral thyroid glands were goiter in gross appearance. Microscopic examination of them disclosed similar pictures. None of them were malignant. Overall, pathologic diagnoses were ectopic thyroid tissue with goitrous change and bilateral thyroid goiter (Figure 4).

DISCUSSION

To our knowledge, goiter can get implanted in the neck after surgery or trauma and then grow. This



A.



B.

FIGURE 4. Histological section. (A) Section of right lateral neck mass revealed encapsulated moiety of ectopic thyroid tissue (white arrow) showing varying sizes of follicles (hematoxylin-eosin stain, original magnification $\times 40$). (B) Section of normally located thyroid goiter showed variable follicles, fibrotic septum and old hemorrhage (hematoxylin-eosin stain, original magnification $\times 40$). [Color figure can be viewed in the online issue, which is available at www.interscience.wiley.com.]

patient did not present any trauma history. Besides, preoperative images and intraoperative findings revealed that bilateral upper poles were intact. Though the detailed information about the previous surgery was not available, we almost can exclude the possibility of postoperative implant and trauma.

Ectopic thyroid tissue is defined as thyroid tissue not located anterolaterally to the second to fourth tracheal cartilages. In 1896, Hickman⁵ first described the lingual thyroid in a newborn who was suffocated 16 hours after birth because of the mass causing upper airway obstruction. The lingual thyroid is the most commonly seen thyroid ectopy and accounts for 90% of reported cases. Other rare sites, such as the mediastinum, lung, porta hepatis, duodenum, esophagus, heart, breast, and intratrachea have been described. Thyroid ectopy in the submandibular space with a coexisting normally located, functioning thyroid gland is extremely rare.^{6–8}

The midline ectopic thyroid along the migration path from foramen cecum to the mediastinum can be explained by the failure of part of the thyroid anlage to descend. Some authors suggest that this phenomenon is likely related to the developmental defects other than abnormal migration of the thyroid bud. The molecular pathology of ectopic thyroid in mouse models has demonstrated that the *Foxe1* mutation is necessary for thyroid migration; however, to date, no known gene is associated with the human ectopic thyroid.⁹ It has been suggested that cells of ultimobranchial bodies can differentiate toward thyroid follicular cells.¹⁰ However, such thyroid follicular cells cannot function as normally developed thyroid follicular cells. Radkowski et al¹¹ and Morgan et al¹² both proposed that hypoglossal duct cysts can be displaced from the midline. Laterally displaced ectopic thyroid tissue is likely related to this phenomenon. The simultaneous identification of dual ectopic thyroid tissue and a normally located functioning thyroid gland is extremely rare. Kuehn et al¹³ reported the only case of dual thyroid ectopy with a normal pretracheal thyroid gland.

From a clinical perspective, any disease that affects normal thyroid glands can also affect ectopic thyroid tissue. Three main entities exist in the differential diagnosis for patients presenting with neck masses: inflammatory diseases, benign neck masses, and malignancies. Currently, fine-needle aspiration cytology is the accepted diagnostic tool for thyroid nodules and has decreased the need for diagnostic thyroidectomy. However, 15% to 20% of all thyroid nodules may be indeter-

minate or suspicious. As submandibular ectopic thyroid tissue is extremely rare, and most neck masses suspected of being ectopic thyroid tissue have been diagnosed as lymph nodes metastasis originating from thyroid papillary cancer, thyroidectomy is typically performed. Therefore, surgical excision is the appropriate therapy and diagnostic modality for suspicious ectopic thyroid tissue.

CONCLUSION

Ectopic submandibular thyroid tissue is an extremely rare entity and poses diagnostic and treatment problems. This report presented an extremely rare case of a slow-growing submandibular ectopic thyroid tissue with coexisting lingual thyroid and normally located functioning thyroid glands. The ectopic thyroid tissue should be considered in treating such cases.

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