# **Ectopic Thyroid Tissue With Hashimoto's Thyroiditis**

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#### ABSTRACT

**Objective:** Ectopic thyroid gland is a rare occurrence with a prevalence of 1 per 100,000 to 300,000 people. Hashimoto's thyroiditis involving ectopic thyroid tissue is particularly unusual. We describe the presentation, workup, surgical management, and brief review of the literature.

**Methods:** Retroactive review of an 83-year-old white female patient record. As a case report, this project was exempt from institutional review board approval.

**Results:** We present a case of ectopic thyroid tissue located in the strap muscles with concurrent Hashimoto's thyroiditis. This tissue initially was believed to represent metastatic follicular thyroid carcinoma.

**Conclusion:** Whenever ectopic thyroid tissue is encountered, the gravest concern is metastatic thyroid cancer. The possibility of benign thyroid tissue should not be excluded even if the thyroid histology initially appears to be malignant in nature.

## INTRODUCTION

Ectopic thyroid tissue is a rare occurrence with a prevalence of about 1 per 100,000 to 300,000 people.<sup>1</sup> It is even more extraordinary if Hashimoto's thyroiditis (HT) is present within that tissue; combined, they represent an ultra rare entity. To date, there are a handful of case reports demonstrating HT within ectopic thyroid tissue, but usually these are found within the normal embryological migration of the thyroid gland. In this case report, we describe a case of HT found in ectopic thyroid tissue within the strap muscles that was initially misinterpreted as metastatic follicular thyroid carcinoma. To our knowledge, the finding of

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extrathyroidal HT located in the strap muscles has not previously been reported.

## **CASE REPORT**

An 83-year-old woman was referred to the Division of Head and Neck Oncology for further management of a newly diagnosed thyroid cancer. She had a past medical history of radiation in 1955 due to tuberculosis; however, radiation type or fields were unknown. Her thyroid history also was significant for radioactive iodine (RAI) ablation in 1998 for Graves' disease, resulting in post-ablation hypothyroidism. During routine examination for hypothyroidism, thyrotropin (TSH) was high at 6.95 uIU/

mL and free thyroxine ( $T_4$ ) was normal at 1.57 ng/dL; an anterior neck mass was noted. An autoimmune work up was not performed. Ultrasound revealed a 5 x 5 x 4 mm hypoechoic nodule of the middle right thyroid lobe and a 4 x 3 x 3 mm calcified nodule of the inferior right thyroid lobe. Prior to detection by her endocrinologist, the patient was unaware of her neck mass and was asymptomatic. She was subsequently referred to an outside otolaryngologist.

A repeat ultrasound was obtained by the outside otolaryngologist, which revealed a  $1.4 \ge 0.6 \ge 1.4$  cm subcutaneous right extrathyroidal neck mass to the right midline near the hyoid bone. Computed tomography (CT) imaging demonstrated an enhancing nodule with a low attenuation center,  $1 \ge 0.7$  cm, embedded in the right strap muscle at the level of the middle right thyroid lobe (Figure 1). Just inferior to that nodule, a similar appearing enhancing nodule also was embedded in the right infrahyoid strap muscle,  $1 \ge 0.7$  cm, at the level of the inferior right thyroid lobe. A third 1.3 cm nodule with 3 mm of central calcification of the right thyroid gland also was found on CT scan imaging. The CT scan was suspicious for metastatic lymph nodes from the right thyroid gland, which were enlarged and had heterogeneous enhancement. Ultrasound guided fine needle aspiration of the 3 nodules revealed extensive infiltration of fibrocollagen stroma by follicular thyroid tissue, which is consistent with metastatic lowgrade follicular cancer per the outside report.

Upon consultation, our pathology department reviewed the histopathologic slides. The superior nodule core biopsy revealed fragments of thyroid follicles with oxyphilic (Hurtle cell) features admixed with dense lymphoid infiltrate with adjacent normal skeletal muscle. On exam, the patient had two easily palpable masses: one measuring 1.5 cm at the level of the hyoid, and the second just about 1 cm below this mass, also to the right of the midline. The right thyroid lobe was firm on palpation. The patient then was taken to the operating room for total thyroidectomy and central dissection for possible metastatic thyroid cancer with a possibility of HT. Metastatic thyroid cancer was in the top differential due to thyroid elements found in ectopic locations. Intra-operatively, two 1.5 cm soft tissue masses were identified: one within the right sternohyoid muscle at the level of the hyoid and one at the level of the thyroid cartilage. Both thyroid lobes had smaller nodules. Surgical pathology demonstrated thyroid tissue within the strap muscles, the existence of normal thyroid tissue outside of the thyroid gland (Figure 2), and HT (Figure 3). The patient was doing well 3 months post surgery and is now scheduled to follow up as needed.

## DISCUSSION

Dr Hakaru Hashimoto is the physician credited for first describing HT, which affects about 2% of the population, making it one of the most common thyroid diseases.<sup>2</sup> It mainly presents in young or middle-aged women, 30 to 50 years old,<sup>3</sup> as a diffuse painful enlargement of the thyroid gland. Hypothyroidism is its most common complication.<sup>4</sup> Up to 40% of patients report a positive family history of thyroid disease. HT is caused by autoimmune destruction, as evident from the presence of antithyroglobulin and antimicrosomal antibodies in the majority of patients.<sup>3</sup> Upon histologic examination of the thyroid glands, there is presence of fibrosis, lymphoid cellular infiltration, larger eosinophilic thyroid cells (Hurthle cells), granulated cytoplasm due to excess mitochondria, and potentially prominent nucleoli.<sup>4</sup> The presence of fibrosis, lymphoid cellular infiltration, and eosinophilic cells was noted in this patient.

Ectopic thyroid tissue is a rare entity occurring in about 1 per 100,000 to 300,000 people, but its prevalence increases to 1 per 4000 to 8000 in people with thyroid disease. In autopsy studies, the prevalence of ectopic thyroid tissue can range from 7% to 10%. In about 70% to 90% of ectopic thyroid tissue, it is the only thyroid tissue present.<sup>1</sup>

Normal embryological development of the thyroid gland starts at the floor of the primitive foregut and migrates caudally to its final position pretracheally, thereby forming the thyroglossal duct.<sup>1</sup> Fusion of 2 anlages makes up the thyroid gland: 2 lateral anlages and a large median endodermal anlage.<sup>5</sup> Figure 1. Axial Computed Tomography (CT) Scan With Contrast of the Head and Neck



Image shows an enhancing nodule,  $1 \times 0.7$  cm (white arrow), at the level of the thyroid cartilage, embedded in the right strap muscle with a central area of low attenuation.

Ectopic tissue is most commonly found laterally in the neck or within the migratory tract, because of a failure to complete descent. The most common location of ectopic thyroid tissue is the tongue around the foramen cecum, known as a lingual thyroid, and this accounts for up to 90% of the cases.1 It can be asymptomatic or present with dysphagia, dysphonia, snoring, stomatolalia, etc. Patients with lingual thyroid most commonly present with hypothyroidism and without orthotopic gland, but less commonly patients may be euthyroid.1 Even more rare, a lateral ectopic gland may be formed if the cells of the lateral anlage do not fuse with the median anlage cells.<sup>1,5</sup> This improper or lack of fusion is commonly found in the submandibular region. This presents as a painless mobile mass in the area of the carotid triangle or in the submandibular area. These ectopic lateral thyroid tissues are mainly right-sided and are more common in women. The patients usually have an orthotopic gland and are euthyroid.<sup>1</sup> Other causes of ectopic lateral thyroid gland include metastasis of thyroid carcinoma, spread of orthotopic thyroid during surgery, and displacement during embryogenesis. Other locations of ectopic thyroid include: intratracheal, intrathoracic, ovarian, gastrointestinal, and adrenal.1

#### CONCLUSION

The aforementioned case reveals extrathyroidal HT found in the strap muscles of an elderly woman with an initial suspicion of a follicular neoplasm. Cassol et al wisely stated that prior to diagnosing thyroid ectopia, careful exclusion of primary thyroid malignancy needs to be performed,<sup>2</sup> which is what the authors in this case pursued prior to diagnosing HT. Whenever there is ectopic thyroid tissue, the convention is for excisional biopsy for

Figure 2. Hashimoto's Thyroiditis of Upper Central Neck Tissue

Abundant follicular lymphoid hyperplasia and oncocytic/Hurthle cell change of the follicles. Nearby skeletal muscle (black solid arrow) and fat (gray solid arrow). H & E stain with 2x (20 x original magnification) magnification.

Figure 3. Ectopic Thyroid Adipose and Soft Tissue

Abundant thyroid follicles (circle) within fibrous tissue (arrow) and fat (white areas) demonstrating existence of ectopic thyroid glandular tissue. H & E stain with 4x (40 x original magnification) magnification.

diagnosis. It would be difficult to differentiate a metastatic thyroid cancer from ectopic thyroid tissue. This case clearly demonstrates how methodical evaluation using ultrasound, CT imaging (to look for extension and location), histopathology, and laboratories are necessary to exclude metastatic thyroid carcinoma. The shortcoming of ultrasound imaging is that it is dependent of the operator skills, thus yielding slightly different results due to interobserver variabilities and interpretation.<sup>6</sup> This case, in particular, was more difficult to diagnose preoperatively due to the patient's history of prior radiation for tuberculosis and RAI ablation, which could have made histopathological diagnosis more obscure. Ultimately, total thyroidectomy was necessary due to the presumptive diagnosis of a follicular neoplasm and metastasis with a small possibility of HT. Also, it is a well-known fact that a "follicular neoplasm" is indeterminate and final diagnosis of malignancy can be confirmed only on final surgical pathology by demonstrating capsule invasion.<sup>7</sup> Currently there is no consensus on management strategies for ectopic thyroid due to the rarity of the condition,<sup>1</sup> thus it is up to the individual surgeon to formulate a proper management plan to treat the patient.

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