A Case of Ectopic Thyroid in Lateral Neck Associated with Graves’ Disease

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Abstract. Thyroid follicles in the lateral position of the neck are usually thought to represent the metastasis of thyroid carcinoma. Here we present a case of a 28-year-old woman with accessory ectopic thyroid associated with Graves’ disease. Despite a history of Graves’ disease poorly controlled with large dose propylthiouracil she was found to be pregnant and artificial abortion was planned. Thyroid scintigraphy was carried out, which indicated an uptake into the region above the left lobe as well as into both lobes of the thyroid gland. In order to control hyperthyroidism and to exclude the possibility of metastasis, total thyroidectomy with tumor resection was performed before the artificial abortion. Pathological examinations of the thyroid gland indicated findings compatible with Graves’ disease. The lateral neck mass was revealed to be composed of nonneoplastic thyroid tissue, showing similar histological findings to those of the goiter, which were consistent with Graves’ disease. Taken together with several previous reports, it appears that there are some cases with lateral ectopic thyroid tissue, whose pathogenetic mechanism remains to be elucidated.

Key words: Ectopic thyroid, Graves’ disease

AN ectopic thyroid usually occurs in the midline as a result of abnormal development [1, 2]. It is thought that ectopic thyroid in the lateral neck almost always represents metastasis from thyroid carcinoma within cervical lymph nodes rather than an embryonic rest [1–3]. We have recently reported a case with multiple aberrant thyroid tissues in the tongue and bilateral cervical lymph nodes [4]. Here we describe a case of a 28-year-old woman with Graves’ disease associated with ectopic thyroid tissue in the lateral cervical region. These cases suggest that thyroid tissue in the lateral cervical region does not always represent malignant tumor, as suggested by other investigators [2, 5–7]. Possible pathogenetic mechanisms of such cases will be discussed.

Case Report

A 28-year-old pregnant woman was referred to our division for the evaluation of hyperthyroidism. She had a history of poorly controlled Graves’ disease despite large dose propylthiouracil (750 mg/day). She had a diffuse elastic soft goiter of 63 mm diameter. Ultrasonography of the thyroid gland revealed diffuse enlargement with hypoechoic level characteristic for treated Graves’ disease and without any formation or calcification. Cytology of fine needle aspiration biopsy showed a remarkable
inflammatory finding mainly with presence of lymphocytes and without any malignant cells. Thyroid function test indicated the presence of hyperthyroidism (free T₄ 2.7 ng/dl, T₃ 18.6 μg/dl, T₃ 273 ng/dl and TSH <0.1 μU/ml) with positive thyrotropin receptor antibody. Artificial abortion was planned and thyroid scintigraphy was carried out with both ⁹⁹ᵐTc and ¹²³I, both of which revealed an uptake into the region above the left lobe as well as into both lobes of the thyroid gland (Fig. 1 left). Cervical computed tomography (Fig. 1 right) showed an enhanced tumor of 1 cm diameter at the same region as the scintigraphy. In order to control hyperthyroidism and to exclude the possibility of metastasis, total thyroidectomy with tumor resection was performed before artificial abortion. Pathological examination of the thyroid gland was performed even 5 mm, which showed findings compatible with Graves' disease. The thyroid gland was diffusely and symmetrically enlarged with smooth capsule. The parenchyma showed a diffuse, uniform hyperplasia, producing numerous papillary projections. The puffs were lined by a single layer of tall columnar cells exhibiting no atypia. The follicles were vacuolated around the edges. There was no histological finding suggesting lymph node, whereas lymphocytic infiltration with well developed germinal centers were seen in the stroma. There was no finding suggesting previous malignancy, such as fibrous scar or psammoma body. The neck mass was composed of nonneoplastic thyroid tissue, showing histological findings similar to those of the thyroid (Fig. 2).

Fig. 1. Thyroid scintigraphy with ⁹⁹ᵐTc (left upper) and ¹²³I (left lower), and computed tomography of the neck (right: the arrow indicates the left neck mass).
An ectopic thyroid is usually considered to occur along the midline from the tongue base to the mediastinum, since, in embryonic morphogenesis, the thyroid gland descends into the neck the foramen cecum of the tongue along the thyroglossal duct [1, 2, 8, 9]. Carcinomas arising in the median ectopic thyroid may constitute a rare entity, but they present diagnostic difficulties for both the clinician and pathologist, the distinction between neoplastic and nonneoplastic ectopic thyroid being difficult to determine. In general, carcinomas arising in the thyroglossal duct are mostly papillary, whereas those in the lingual thyroid are not [1, 2, 9-11]. In contrast, ectopic thyroid of the lateral neck almost always has been considered to represent a metastasis from the thyroid carcinoma [1-3], and only rarely a primary carcinoma arising in the ectopic thyroid [12]. However, several cases have been reported in which normal thyroid tissue was found in the lateral cervical regions [2, 5-7, 13]. Recently we reported a 37-year-old woman who had multiple aberrant thyroid tissues in the tongue as well as bilateral cervical lymph nodes. The histological findings indicated a diagnosis of adenomatous goiter, and the molecular analysis using X-inactivation of human androgen receptor gene revealed that the lymph nodes were of polyclonal origin, confirming their nonneoplastic nature [4].

In the present case, histological examinations of both the goiter and the lateral neck mass indicated a diagnosis of Graves' disease without any evidence of malignancy. Unfortunately the results of X-inactivation method using human androgen receptor or phosphoglycerate kinase-1 gene was not informative, since neither of the DNA markers was heterogeneous (data not shown). Therefore the possibility of primary occult thyroid carcinoma arising from the ectopic thyroid, especially of follicular carcinoma [10] or of the follicular variant of papillary carcinoma with hyperthyroidism [14], should be considered. However, that possibility seems remote considering the fact that the lateral neck mass did not show any nuclear or cellular features of malignancy, but showed totally similar findings to those of the thyroid gland with typical features of Graves' disease. Further, the possibility of metastasis from the occult primary carcinoma in the thyroid, as suggested in the carcinoma in the median ectopic thyroid [9], was excluded in this case, since the histological specimens

**Fig. 2.** Photomicrograph of the specimen obtained from the thyroid (left) and the left neck mass (right) (H–E stain: original magnification × 100).
obtained from total thyroidectomy revealed no evidence of malignancy. Furthermore, the lateral mass was connected to the left lobe of the thyroid with tubular structures. Therefore, the possibility cannot be excluded that the lateral thyroid tissue was not a true ectopic one, but was derived from the large goiter due to Graves’ disease, which had shown shrinkage with the residual tissue in the lateral region. There are several possible mechanisms to explain the etiology of ectopic thyroid in the present case. First, as described above, the lateral thyroid tissue may not be a true ectopic one, but a remnant derived from Graves’ disease, which had shown shrinkage. Second, some developmental anomaly has occurred, as in thyroid hemiagenesis [15]. However, an abnormally developing lateral lobe is generally considered to give rise only to C cells, but not follicular cells [7, 12, 16, 17]. Furthermore, there was no finding which suggested the remnants of ultimobranchial body, such as abundant C cells or solid cell nest. Third, the ectopic thyroid tissue may be derived from the thyroglossal duct, considering that its position was relatively near to the midline region. The histological examinations may support the first hypothesis, i.e., the pathological findings of the mass compatible with Graves’ disease and the presence of tubular connections of the lateral mass and the left lobe. However, the exact etiological mechanism of the condition remains to be elucidated.

In summary we reported a case of Graves’ disease with ectopic thyroid tissue in the lateral region, whose pathogenetic mechanism remains to be elucidated.

References