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Case Report

Papillary Thyroid Carcinoma Arising from a Median Ectopic Thyroid with No Thyroglossal Duct Remnant

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Thyroid carcinomas arising from ectopic thyroid tissue are uncommon; most of them arise from thyroid tissue in thyroglossal cysts. A rare case of a 66-year-old woman with papillary thyroid carcinoma arising from median ectopic thyroid tissue lacking a thyroglossal duct remnant is reported. The tumor was resected by Sistrunk's procedure, and the patient's postoperative course was good.

Key words: ectopic thyroid, papillary thyroid carcinoma, thyroglossal duct remnant

Ectopic thyroid tissue is a rare entity resulting from developmental defects at early stages of thyroid gland embryogenesis during its passage from the floor of the primitive foregut to its final pre-tracheal position [1]. Normally, by the 7th embryonic week, thyroid tissue comes to lie anterior to the developing trachea, and, by then, the thyroglossal duct has begun to atrophy, with the original pharyngeal connection of the duct identified at the apex of the sulcus terminalis on the dorsum of the tongue, the foramen cecum [2]. About 60% of thyroglossal duct remnants are located between the hyoid bone and the thyroid cartilage, and the majority of thyroglossal carcinomas occur in this region [2, 3]. We present a rare case of papillary thyroid carcinoma (PTC) arising from an ectopic thyroid in front of the thyroid cartilage lacking a thyroglossal duct remnant.

Case Report

A 66-year-old Japanese woman was referred to our

department for further evaluation of a median anterior neck mass. She complained of anterior neck discomfort that had lasted 1 month, but she had no significant past medical history. No abnormalities were detected in the larynx or hypopharynx on magnifying electroendoscopy. Examination of the neck revealed a firm mass with an irregular edge and good mobility. While free thyroxin (1.18 ng/dl), thyroglobulin antibodies (11 IU/ml), and thyroid peroxidase antibodies (5 IU/ml) levels were all normal, thyroid-stimulating hormone (4.78 μ IU/ml) and thyroglobulin (62.9 ng/ml) levels were slightly elevated.

Ultrasonography (US) revealed a solid subcutaneous mass (18.5 mm \times 18.1 mm \times 16.5 mm) in front of the thyroid cartilage (Fig. 1) and a normally positioned thyroid gland. The thyroid gland had scattered minute cystic lesions, but no solid mass. Computed tomography showed a uniformly enhanced mass with a clear border in front of the thyroid cartilage. The dorsal part of the mass showed a high density equal to that of thyroid tissue (Fig. 2). By fine needle aspiration, many cell clusters were obtained. The cells were cuboidal epithelial cells and arranged in sheets or in papillary configurations. Some of them had nuclear grooves or intranuclear cytoplasmic inclusions. These

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cytological features are characteristic of PTC. Three differential diagnoses were considered: ectopic thyroid carcinoma, lymph node metastasis from PTC of the orthotopic gland, and pyramidal lobe carcinoma. No lymph node metastasis was detected by preoperative US examination. Surgery was performed. The mass was found on the sternohyoid muscle, and it had no continuity with the orthotopic thyroid gland. Although no thyroglossal duct remnant was observed during the surgery, the mass was completely dissected by Sistrunk's procedure, including the middle portion of the hyoid bone (Fig. 3). Thyroidectomy was not performed. Intraoperative histopathological examina-

tion revealed the existence of ectopic thyroid tissue in addition to PTC, and postoperative histopathological study found no thyroglossal duct tissue. The final diagnosis was PTC arising from ectopic thyroid tissue (Fig. 4). Thyroid function did not deteriorate after the surgery. The levels of thyroid-stimulating hormone and thyroglobulin were $4.12\mu\text{IU}/\text{ml}$ and $35.1\text{ ng}/\text{ml}$ at 7 months after the surgery.

Discussion

The most frequent location of ectopic thyroid tissue is at the base of tongue, accounting for about 90% of reported cases [4]. Apart from the region of the foramen cecum, ectopic thyroid tissue has been

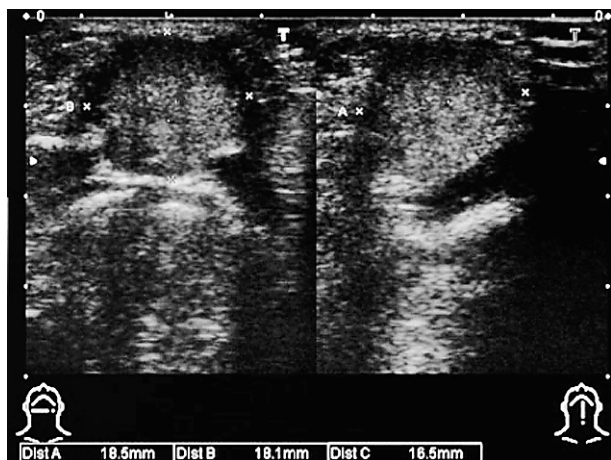


Fig. 1 Ultrasonography of the neck region. A subcutaneous solid mass ($18.5\text{ mm} \times 18.1\text{ mm} \times 16.5\text{ mm}$) is present on the thyroid cartilage.

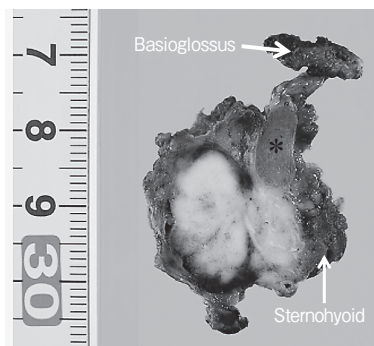


Fig. 3 Isolated preparation. The tumor was dissected with adipose tissue and the basioglossus and sternohyoid muscles. There was ectopic normal thyroid tissue (*) on the dorsal surface of the tumor.

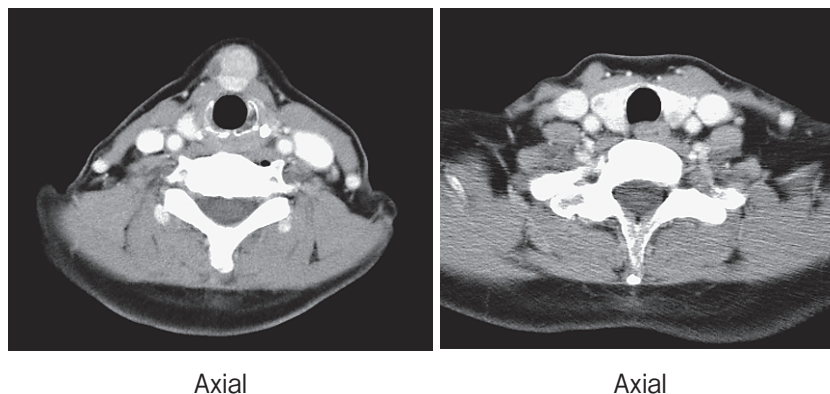


Fig. 2 Computed tomography with 5-mm slice thickness shows the tumor and the orthotopic thyroid gland. The right picture is taken at 4 slices below left picture. A uniformly enhanced mass with a clear border is seen on the thyroid cartilage, and an area with high density equal to that of thyroid tissue is seen on the dorsal surface of the mass. The mass is not continuous with the orthotopic thyroid gland.

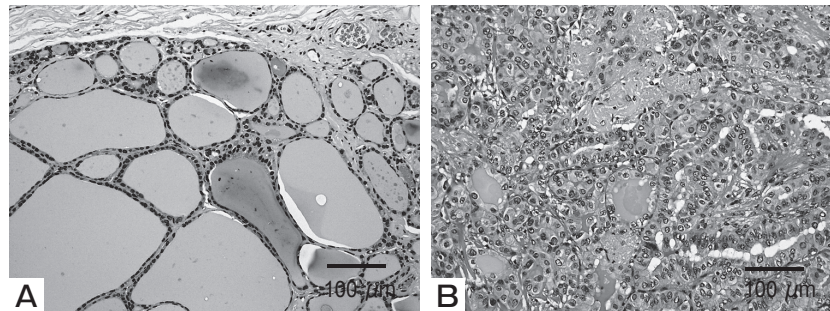


Fig. 4 Histopathological examination showed normal thyroid tissue **A** and a papillary carcinoma **B** (hematoxylin and eosin stain).

described in numerous other sites between the base of the tongue and its final pre-tracheal position, as well as in the mediastinum and distant subdiaphragmatic areas [1]. It may be difficult to distinguish a thyroglossal duct cyst clinically from an ectopic thyroid gland [5], and inadvertent extirpation of ectopic thyroid tissue is sometimes performed, leading to hypothyroidism [6]. On the other hand, it is estimated that 35% to 70% of thyroglossal duct cysts may contain thyroid tissue in their walls [7]. Carcinomas arising from a thyroglossal duct cyst are rare (1% of thyroglossal duct cyst cases), but about 80% of them are PTC, which should behave in the same way as PTC in the orthotopic gland [8, 9]. Furthermore, thyroid carcinoma arising from ectopic thyroid tissue lacking a thyroglossal duct is rare, and only a few cases of this condition have been reported in English or Japanese [10, 11]. A case of an ectopic thyroid nodule whose location was very similar to that in the present case was reported as ectopic thyroid tissue in a thyroglossal duct cyst, but the existence of a thyroglossal duct remnant was not confirmed surgically [12]. In the authors' experience, such cases occasionally present in which the preoperative diagnosis is thyroglossal duct cyst, but detection of a thyroglossal duct remnant is difficult even surgically. Therefore, it might be inappropriate to consider all cases with ectopic thyroid tissue lacking a thyroglossal duct as truly lacking a duct remnant. However, in the present study, the lack of a thyroglossal duct was confirmed by histopathological examination.

The clinician must also distinguish between ectopic thyroid carcinomas and metastases of thyroid carcinoma of the orthotopic gland that have totally replaced a lymph node [1, 8, 13]. Virginia A. LiVolsi *et al.* reported that a total thyroidectomy might not neces-

sarily be indicated as a routine procedure following the finding of a papillary carcinoma in the thyroid region, especially if normal thyroid tissue or thyroglossal duct remnants are found [14]. If the thyroglossal duct cyst is uncertain, physicians should check preoperatively whether there is normal thyroid tissue with the tumor. Since the thyroid gland was in the normal site in the present case, the possibility of metastasis from an occult PTC could not be completely ruled out. However, sonographic evaluation revealed no solid mass in the orthotopic gland, and CT showed a high density equal to that of thyroid tissue at the dorsal part of the tumor. Thus, we considered that the PTC in the present case originated from ectopic thyroid tissue. Since this case was low-risk according to the PTC classification [15], performing additional total thyroidectomy followed by radioactive iodine therapy seemed excessive. Although no thyroglossal duct remnant was observed during the surgery, we could not completely rule out the possibility of ectopic thyroid carcinoma with thyroglossal duct remnant. Therefore, the mass was dissected by Sistrunk's procedure, but long-term follow-up will be necessary. In 1928, Sistrunk described a procedure to excise thyroglossal duct cysts [8]. This procedure included resection of the cyst and tract, extending to the foramen cecum at the base of the tongue in continuity with the midportion of the hyoid bone. We did not perform cervical lymph node dissection in this case. Our institution has been performing routine preoperative ultrasound examinations, and prophylactic neck dissection for patients without preoperatively positive nodes in the lateral compartment is not usually performed [16].

Finally, we must consider the possibility that the ectopic thyroid may present as the only functional

thyroid tissue despite the existence of an orthotopic thyroid gland [17]. Checking postoperative thyroid function is important. In the present case, since the preoperative serum TSH level was slightly elevated, more attention to the postoperative thyroid function will be needed. Although we had no choice but to perform surgery this time, careful preoperative informed consent should be obtained from the patient in all such cases.

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