



Preoperative Diagnosis of Papillary Carcinoma Arising from Thyroglossal Duct Cyst: Two Case Reports and Literature Review

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Abstract

Thyroglossal Duct Cyst (TGDC) is a common disease that rarely develops into carcinoma. We report two cases of papillary carcinoma arising from TGDC diagnosed preoperatively.

Case 1: A 36-year-old woman presented with a painless mass in the midline of the neck. Computed Tomography (CT) showed an 18 mm mass lesion in the area surrounded by the hyoid bone and thyroid cartilage and bilateral cervical lymph node enlargements. The pathological findings were papillary carcinoma with bilateral lymph node metastases.

Case 2: A 44-year-old woman presented with a painless mass in the midline of the neck. CT showed a cystic lesion with a 37 mm calcification and a partial contrast effect in the middle of the hyoid bone. The pathological findings were papillary carcinoma without lymph node metastases. Both lesions were definitively diagnosed by Fine Needle Aspiration (FNA). Both patients were followed up without adjuvant therapy and had good courses without recurrence at 31 and 29 months after surgery. The possibility of thyroglossal duct cancer should be kept in mind if preoperative imaging shows findings that are not typical of TGDC, such as thickening or irregularities of the cyst wall or solid lesions within the cyst and calcification. Preoperative ultrasound-guided FNA is effective for developing an appropriate surgical plan if we suspect malignancy.

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Introduction

A Thyroglossal Duct Cyst (TGDC) is a cervical tumor that develops from the remnant of the thyroid duct during the embryonic period. It is generally known as a benign disease. TGDCs often have normal thyroid tissue [1] but rarely develop into carcinomas. A definitive diagnosis of malignancy is often made after surgery and is extremely rarely made before surgery. We report two cases of preoperatively diagnosed papillary carcinoma arising from the thyroglossal duct that were appropriately treated with good progress.

Case Series

Case 1

A 36-year-old female patient presented with a painless mass in the midline of the neck. A tumor about 20 mm in diameter was palpable. The tumor was an elastic, hard, movable mass lesion. Laboratory results showed only a slight elevation of C-reactive protein (0.75 mg/dl) and no abnormalities in peripheral blood images, liver function, and kidney function. Tumor markers and thyroid function were within normal ranges. Computed Tomography (CT) revealed a 1.8 cm × 1.6 cm × 2 cm occupied lesion between the hyoid bone and the thyroid cartilage (Figure 1). The margin was accompanied by calcification. The tumor margin was well circumscribed, and involvement of the attached organs and bone destruction were not observed. Right submandibular and bilateral lower internal deep cervical lymph node enlargement (approximately 11 mm to 14 mm in diameter) was observed. Ultrasound-guided Fine Needle Aspiration (FNA) revealed papillary carcinoma (Figure 2). Operation was performed under the diagnosis of papillary carcinoma arising from TGDC.

Intraoperative findings: Tumorectomy combined with excision of the body of the hyoid bone was performed with sufficient surgical margins. Bilateral modified neck dissection was also performed. The thyroid gland was preserved, because there was no continuity with the pyramidal lobe. Both sides of the sternocleidomastoid muscle, the internal jugular vein, the hypoglossal nerve,

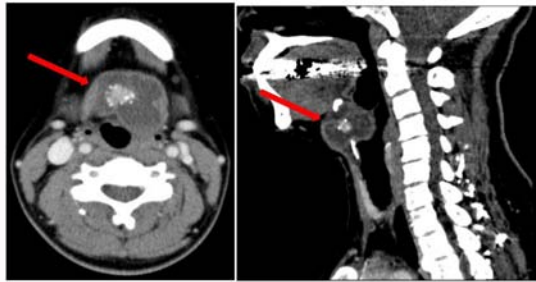


Figure 1: CT revealed a 1.8 cm x 1.6 cm x 2 cm occupied lesion (arrows) between the hyoid bone and the thyroid cartilage. The margin was accompanied by calcification. The tumor margin was well circumscribed, and involvement of the attached organs and bone destruction were not observed. Right submandibular and bilateral lower internal deep cervical lymph node enlargement (approximately 11 mm to 14 mm) was observed.

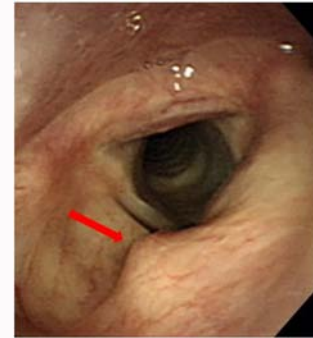


Figure 4: Laryngeal fiber findings. Exclusion of the epiglottis due to compression by the mass (arrow) was observed. No recurrent nerve palsy was noted.

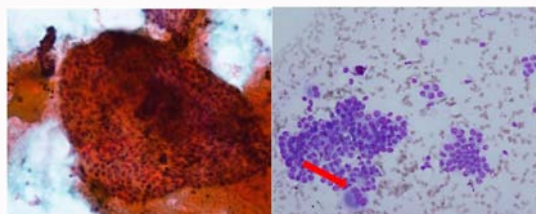


Figure 2: Left: Pap staining. Objective x40. Swollen, disorganized atypical cells were observed in the clumps. Right: Giemsa staining. Objective x40. Chromatin was increased, and cells with nuclear grooves (arrow) and intranuclear inclusions were observed.

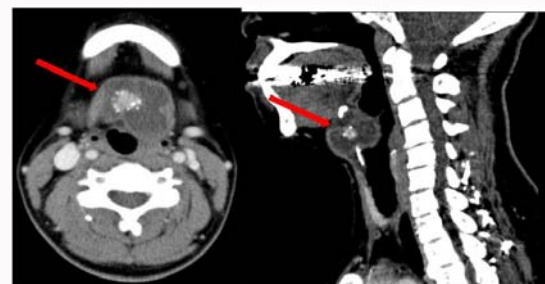


Figure 5: CT showed a 37 mm cystic lesion between the hyoid bones. The mass was depressing the epiglottis (arrow). It was accompanied by calcifications. There was no significant lymph node swelling.

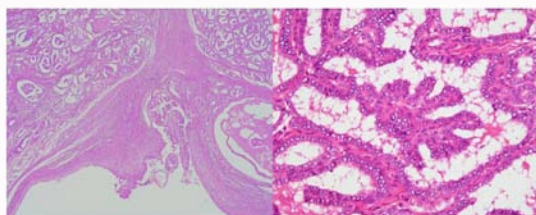


Figure 3: Left: Hematoxylin and eosin (HE). Objective x40. Cystic lesions lining the squamous epithelium are formed. Right: HE. Objective x40. Proliferation and invasion of papillary carcinoma are observed in the cystic cavity.

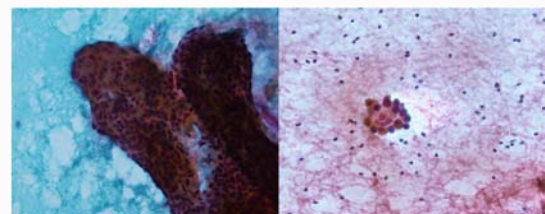


Figure 6: Left: Pap staining. Objective x40. Large aggregates with high nuclear ratio and strong cell binding in the foam cells are seen. Right: Pap staining. Objective x40. Chromatin was granular and abundant. Nuclear grooves and nuclear inclusions were observed in part. A papillary clump and a few calcifications were also noted, and papillary carcinoma arising from the thyroid tissue was strongly suspected.

the phrenic nerve, the accessory nerve, the vagal nerve, and the submandibular glands were preserved.

Pathological findings: Proliferation and invasion of papillary carcinoma were observed in the cystic cavity. Lymph node metastases were found in both cervical lymph nodes (Figure 3). Invasion of the surrounding tissue was not observed, and the surgical stump was negative for cancer.

Postoperative course: The postoperative course was uneventful, and the patient was discharged on postoperative day 7. The patient was alive without recurrence 31 months after surgery, without adjuvant therapy.

Case 2

A 44-year-old female patient presented with neck swelling. A tumor about 30 mm in diameter was palpable on the midline of the neck. The tumor was elastic, hard, and movable. The cervical lymph nodes were not swollen. There was no palpable mass in the thyroid gland. Laryngeal fiber showed exclusion of the epiglottis due

to compression by the mass. The recurrent nerve was not paralyzed (Figure 4). Laboratory results showed a slight decrease in hemoglobin (7.7 g/dl), and no abnormalities were found in peripheral blood images, liver function, or kidney function. Tumor markers and thyroid function were within normal ranges.

CT revealed a 37 mm cystic lesion between the hyoid bones. The mass was depressing the epiglottis. It was accompanied by calcifications (Figure 5). There was no significant lymph node swelling. FNA revealed papillary carcinoma (Figure 6). Operation was performed under the diagnosis of papillary carcinoma arising from TGDC.

Intraoperative findings: The tumor had invaded to the inside of the larynx from the upper end of the thyroid cartilage, but it could be removed without breaking into the pharyngeal cavity. Bilateral modified neck dissection was also performed. The pyramidal lobe

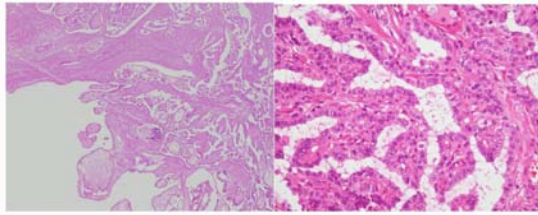


Figure 7: Left: Hematoxylin and eosin (HE). Objective x4. Covering with cell clumps consisting of irregular and dense proliferation of heteromorphic cells could be observed in cysts. Right: HE. Objective x40. Atypical cells were associated with intranuclear inclusions. Similar heterotypic cells were also growing papillary within the cyst wall.

was found to be continuous with the tumor and therefore, total thyroidectomy was performed. Both sides of the submandibular glands, the internal jugular vein, the sternocleidomastoid muscle, the accessory nerve, the vagal nerve, and the sublingual nerve were preserved.

Pathological findings: Proliferation and invasion of cancer cells similar to papillary thyroid carcinoma were observed in the cystic cavity. The dissected lymph nodes were not involved. No invasion of the surrounding tissues was found, and the surgical stump was negative for cancer (Figure 7).

Postoperative course: Mild laryngeal edema and dysphagia were noted postoperatively. The symptoms gradually improved, and the patient was discharged on postoperative day 21. The patient was alive without recurrence 29 months after surgery, without adjuvant therapy.

Discussion

The thyroglossal duct is an epithelial lumen that connects the foramen cecum of the tongue to the thyroid gland at the time when the thyroid gland descends from the embryonic pharynx to the normal position in gestational weeks 4 to 7. It usually shrinks and disappears after the seventh week of pregnancy. It has been reported in about 7% of adults [2].

Only 1.6% to 1.9% of TGDCs are associated with malignant tumors [1,3,4]. Li Volsi et al. [1] reported that ectopic thyroid tissue was found in the cyst wall in more than 60% of TGDCs when the inside of the cyst was examined in detail. It is possible that the cancers develop from the thyroid gland remnant.

Thyroglossal duct cancer was first reported by Brentano et al. [5] in 1911, and since then, only about 200 cases have been reported [6]. Our review of the literature found only 86 reported cases from Japan. According to van Vuuren et al., the average age at diagnosis was 40.0 years (range, 4 to 84 years), and the ratio of male to female cases was 1:1.7 [7]. In Japan, the average age at diagnosis was 44.2 years (range, 11 to 77 years), and the ratio of male to female cases was also 1:1.7.

Pathologically, papillary carcinoma is the most common type, accounting for 79.7% of thyroglossal duct cancers according to van Vuuren et al. [8]. In Japan, papillary carcinoma was reported in 76 of 86 cases of thyroglossal duct cancer (88.3%), squamous cell carcinoma in 5 cases (5.8%), follicular carcinoma in 2 cases (2.3%), papillary follicular carcinoma in 1 case (1.1%), mucinous adenocarcinoma in 1 case (1.1%), and MALT lymphoma in 1 case (1.1%) [8]. In Japan, papillary carcinoma also accounted for most cases of thyroglossal duct cancer. The absence of medullary carcinoma is thought to be

due to the absence of parafollicular cells that are derived from the ultimobranchial body [9].

In most cases, a definitive diagnosis can be made after surgery for TGDC. It is extremely difficult to make an accurate diagnosis before surgery. Even in cases in which FNA is performed, often only residual thyroid tissues or cyst fluids are aspirated if the cancer is confined to a part of the remaining tissue of the thyroglossal duct. The diagnosis of malignancy can be made in only 50% to 60% of all resected tumors [10,11]. Ultrasound examination and imaging methods such as CT and magnetic resonance imaging are generally used. Findings suggesting the development of cancer include thickening and irregularities of the cyst wall and the presence of solid lesions inside the cyst wall. The presence of calcification has been reported to be especially important [12]. In both of our cases, accurate results were obtained by FNA from calcified lesions under ultrasound guidance. We were able to select an appropriate surgical procedure because a diagnosis of malignancy was obtained before surgery.

The Sistrunk method is generally used in surgery for thyroglossal duct cancer [13]. The intraoperative findings of case 2 suggested continuity between the pyramidal lobe and the tumor, and therefore, total thyroidectomy was also performed. However, no malignancy was found in the thyroid.

In 17.1% of thyroglossal duct cancers, the cancer invades the surrounding tissues beyond the cyst wall, and the lymph nodes are involved in 14.6% of cancers [8]. In such cases, neck dissection must be performed at the same time as surgery for thyroglossal duct cancer. Bilateral modified neck dissection was performed in our two cases. Case 1 had bilateral lymph node metastases. Neck dissection should be taken into consideration in cases with malignant findings from FNA performed before surgery or cervical lymph node swelling from imaging studies.

In the 86 cases reported from Japan, the prognoses were satisfactory. There were only two deaths from tumors (one from squamous cell carcinoma and one from follicular carcinoma). Death from tumor has not been reported for papillary carcinoma.

Because the tumors were completely excised, both patients in our study were followed up without adjuvant therapy, and both were alive and well without recurrence at 31 and 29 months after surgery. We report two cases of papillary carcinoma arising from a TGDC diagnosed before surgery by imaging and ultrasound-guided FNA. A good course was obtained in both patients. The possibility of thyroglossal duct cancer should be kept in mind if preoperative imaging shows findings that are not typical of TGDCs, such as thickening or irregularities of the cyst wall or solid lesions within the cyst. In particular, malignancy should be strongly suspected in cases involving solid or calcified lesions.

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