Case Report

METASTATIC PAPILLARY THYROID CARCINOMA ARISING FROM THYROGLOSSAL DUCT CYSTS

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ABSTRACT

Objective: We report a rare case of a patient who developed papillary thyroid carcinoma (PTC) arising from thyroglossal duct cysts (TGDCs) with lateral lymph node metastasis who had a normal thyroid gland and benign central compartment lymph nodes.

Methods: We present a case with metastatic PTC arising from TGDC together with a brief review of the relevant literature.

Results: A 38-year-old man patient was admitted with a cystic lesion in the midline of his neck. There was also an easily palpable lymph node on the left neck, which was suspected of being metastatic PTC on ultrasonography (USG). There was no nodule in the thyroid gland. A fine-needle aspiration cytology of the lymph node was positive for metastases of PTC. We performed an excision of the cystic lesion and total thyroidectomy with the central and left neck lymph node dissection. The cystic lesion pathology was reported as PTC, 10 mm in size. There was a metastatic lymph node in the lateral left neck compartment (regions III-IV), 7 cm in diameter, without central lymph node involvement. Radioactive iodine ablation therapy with 150 mCi of ^{131}I was given after the surgery. The patient is in remission for PTC 2 years after his surgery, and he is currently receiving levothyroxine suppression therapy.

Conclusion: While evaluating a TGDC, a detailed neck USG should be performed in all cases. Furthermore, a biopsy should be conducted of suspected lymph nodes before planning the surgery. **(AACE Clinical Case Rep. 2016;2:e325-e328)**

Abbreviations:
DTC = differentiated thyroid cancer; **FNA** = fine-needle aspiration; **MRI** = magnetic resonance imaging; **PTC** = papillary thyroid carcinoma; **RAI** = radioactive iodine ablation; **TGD** = thyroglossal duct; **TGDC** = thyroglossal duct cyst; **USG** = ultrasonography

INTRODUCTION

Thyroglossal duct cysts (TGDCs) are the most common form of congenital cysts on the neck. Thyroglossal duct (TGD)-associated malignancy is rare (about 1%), and the majority of cases of papillary thyroid carcinoma (PTC) are thyroid-gland type (1-3). Metastasis to the regional lymph nodes has been reported in 7.7 to 12.9% of TGD-associated PTCs, which is much less frequently than primary papillary carcinomas of the thyroid gland (4).

We report a case of PTC arising from TGDC with lateral lymph node metastasis; the patient had a normal thyroid gland and benign central compartment lymph nodes.

CASE REPORT

A 38-year-old man was admitted with complaints of swelling in his neck. On physical examination, the thyroid gland was not palpable, and a painless mass approximately 2 cm in diameter was palpated in the midline of the neck.

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Thyroid function tests were normal, and thyroid autoantibodies were negative. We observed a cystic lesion approximately 16 × 32 mm in size on ultrasonography (USG). The thyroid gland ultrasound was normal. We also found a lymph node, measuring roughly 55 × 25 mm in size, on the left neck compartment (regions III-IV), which was suspected to be metastasis of PTC. This solid lesion with cystic components was 53 × 23 × 16 mm in size and hyperintense on T2-weighted sequences on neck magnetic resonance imaging (MRI) (Fig. 1). In the first evaluation of the MRI imaging, this lesion was thought to be hemangioma or lymphangioma. However, experienced radiologists who re-examined the MRI images decided that the lesion was a metastatic lymph node.

Fine-needle aspiration (FNA) cytology of the lymph node was positive for metastases of PTC. We accordingly performed TGDC excision and total thyroidectomy with central and left lateral neck lymph node dissection. The TGDC specimen pathology was reported as PTC, 10 mm in size (Fig. 2). There was a metastatic lymph node in the lateral left neck compartment, 7 cm in diameter, without central lymph node involvement. After the operation, there was no significant residual thyroid tissue or suspicious cervical lymph nodes based on USG imaging of the neck.

Radioactive iodine (RAI) ablation therapy with 150 mCi of ^{131}I was given after the operation; there was minimal residual focal uptake in the left side of the thyroid region, but no evidence of distant metastases seen on the post-therapy scan. After the RAI ablation, 150-μg levothyroxine therapy was initiated, and the patient was monitored closely. An ^{131}I whole-body scan was performed 6 months after the RAI ablation therapy; we found no evidence of abnormal iodine uptake. Serum thyroglobulin level was <0.2 ng/mL in the second year of follow-up. The patient has been in remission for the 2 years that have elapsed since his surgery.

DISCUSSION

TGDCs are the most common form of congenital cysts on the neck. These are cysts of epithelial remnants of the thyroglossal tract and are typically observed as midline neck masses. TGDCs are often within 2 cm of the midline (usually in the left), and they are typically associated with the hyoid bone (5). Although most patients with TGDCs are children or adolescents, up to one-third of patients are age 20 or older. Histologically, TGDCs contain squamous or pseudostratified ciliated columnar epithelium and ectopic thyroid gland tissue in the duct wall. TGD-associated malignancy is rare (about 1%), and the majority of cases are PTC (6). Two histopathologic and diagnostic criteria for primary carcinoma of the TGDC have been suggested by Widström et al (7): localization of the carcinoma to a clearly demonstrable TGD and the absence of carcinoma on histopathologic examination of the thyroid gland. Approximately 260 cases of papillary carcinomas in TGDC have been reported in the literature since the first report in 1911; the majority of these reports consist of single case reports and small series. According to the literature, 88% of TGD-associated malignancies are PTC, 6% are squamous cell carcinoma, and the rest include Hürthle cell, follicular, and anaplastic carcinomas (4,8-10). Medullary thyroid carcinoma has not been reported from TGDCs (4). Metastasis to regional lymph nodes has been reported in 7.7 to 12.9% of TGD-associated PTCs (4). However, lymph node metastasis has been reported in 40.5 to 75% of patients, particularly if a concomitant PTC in the thyroid gland is present (10,11). In a series of 16 patients characterized by PTC arising in a TGDC who underwent neck dissection of the central and/or lateral compartments, lymph node metastases were found in 75% (12 of 16) of individuals. Approximately half of these cases also had a small foci of papillary cancer in their thyroid glands. Forty percent

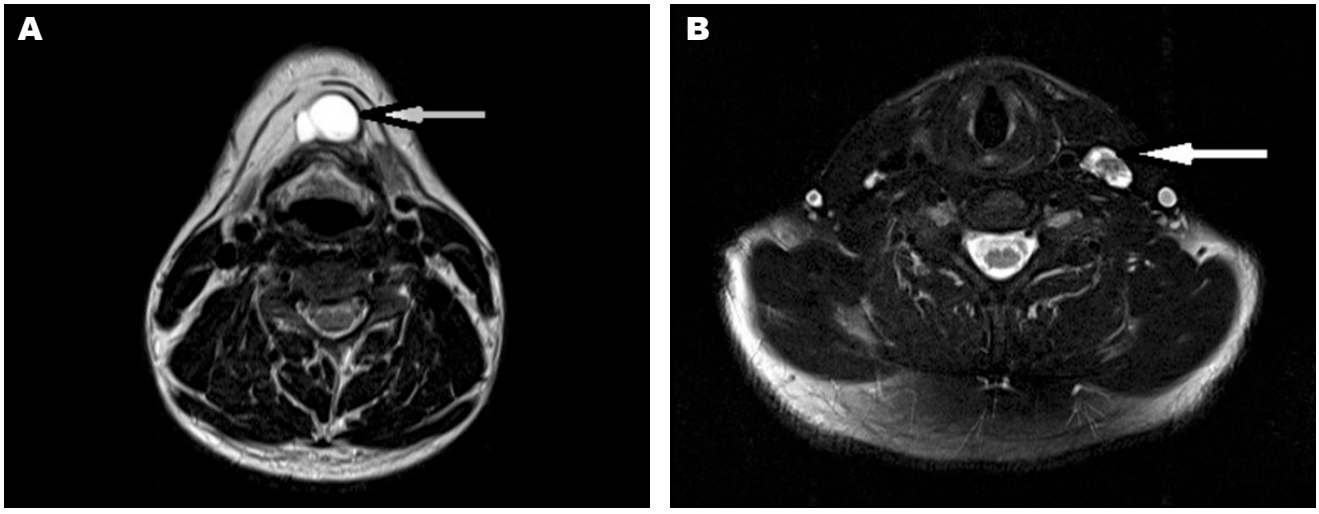


Fig. 1. *A*, Thyroglossal duct cysts, T2-weighted sequences on neck magnetic resonance imaging. *B*, Metastatic lymph node, T2-weighted sequences on neck magnetic resonance imaging.

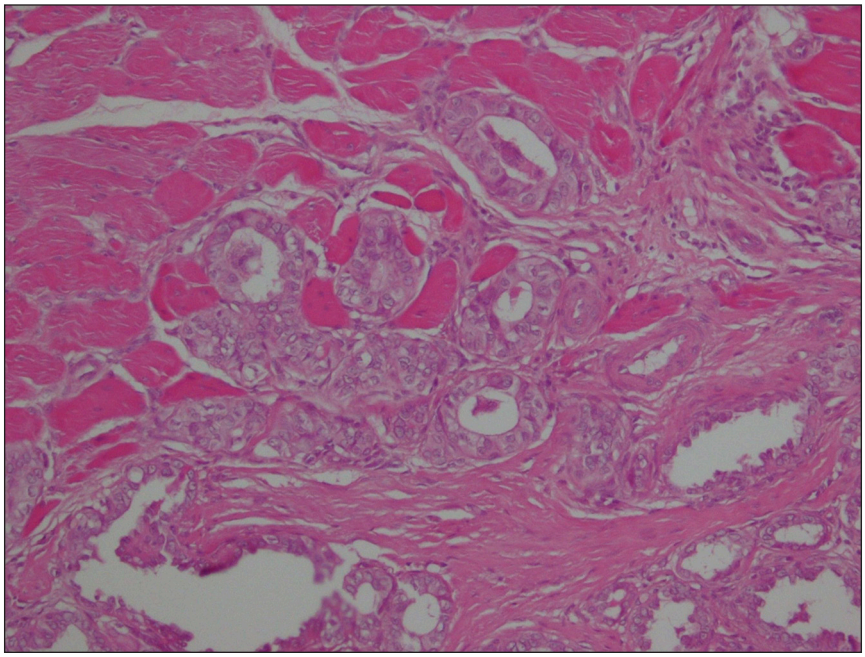


Fig. 2. Papillary carcinoma within a thyroglossal duct cyst, thyroid papillary carcinoma infiltrating muscle (hematoxylin and eosin staining, magnification ×200).

of cases (6 of 15) exhibited lateral lymph node metastasis without central lymph node metastasis (12). In another single-center retrospective study of 26 patients with differentiated thyroid cancer (DTC) in TGDC, a concomitant PTC in the thyroid gland was found in 16 out of 26 cases (61.5%), and it was multifocal in 8 of 16 cases (50%). The central compartment was involved in 6 patients (35.3%, all with concomitant carcinoma in the thyroid gland), and the laterocervical compartments was involved in 10 patients (58.8%). Finally, the submental compartment was involved in 4 patients (23.5%) (11). Distant metastasis is rare (1).

FNA is often used to diagnose TGDCs, but it has only a moderate sensitivity. Its cytomorphologic features include colloids, macrophages, lymphocytes, neutrophils, and ciliated columnar cells, but these features are not unique to TGDCs. Although cytology has moderate sensitivity, it is useful for excluding other diagnoses with an appropriate clinicoradiologic examination (13).

USG is an extremely useful modality for thyroid glands since it reveals the cystic nature of a TGDC. However, USG does not provide information about the relationship with surrounding structures including the hyoid bone. As a result, contrast enhanced computed tomography or MRI are more valuable than USG for determining the relationship between the TGDC and other structures in the neck (14).

The standard treatment for a TGDC is Sistrunk procedure: resection of the duct to the base of the tongue and the central portion of the hyoid bone (15). When lymph node metastases are detected pre-operatively, a total thyroidectomy and regional neck dissection are recommended in addition to the Sistrunk procedure for TGD-associated PTCs (15). Suppressive hormone therapy and radioactive iodine are recommended for these patients (15). However, according to some researchers, ultrasound screening of the thyroid might be a reasonable option for surveillance of the patients with only small TGDC-associated PTCs without lymph node involvement, instead of total thyroidectomy. There is no evidence-based consensus that total thyroidectomy is necessary for the optimal treatment of these low-risk patients (16,6).

Our case, which satisfied the diagnostic criteria of TGD-associated PTC (7), is rare due to its having lateral lymph node metastasis without central lymph node and thyroid gland involvement.

CONCLUSION

In conclusion, while evaluating a TGDC, a detailed neck USG should be performed in all cases, and a biopsy should be conducted of suspected lymph nodes prior to planning the surgery. Patients with lymph node involvement (confirmed by FNA) require both TGDC resection and total thyroidectomy with selective LN dissection. A careful histologic examination of both the surgical specimen and associated lymph nodes and the thyroid gland is mandatory. Subsequent management should adhere to current DTC guidelines.

DISCLOSURE

The authors have no multiplicity of interest to disclose.

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