INTRODUCTION

Thyroglossal duct cysts (TGDC) are the most common form of congenital cysts on the neck. The incidence of thyroid papillary carcinoma in thyroglossal duct cyst is less than 1%. Regional lymph node metastasis is usually seen in cases who have intrathyroidal tumor.

We will report a case of thyroid papillary carcinoma arising from thyroglossal duct cyst with lateral lymph node metastasis who have a normal thyroid gland and benign central compartment lymph nodes.

CASE REPORT

A 38-year-old male patient was admitted with the complaint of swelling in his neck. On physical examination, the thyroid was not palpable and a painless mass about 2 cm in diameter was palpated in the midline of the neck. Thyroid function tests were normal and thyroid autoantibodies were negative. It was a cystic lesion (thyroglossal duct cyst?) and 16x32 mm in size on ultrasonography (USG). The thyroid gland ultrasound was normal. There was also a big mass (lymph node?) on the left neck extending up from the upper to the lower jugular chain, measured about 55x25 mm in size, containing millimetric calcifications and central and peripheral vascularization which was suspected to be metastasis of papillary thyroid carcinoma. This solid lesion with cystic components was 53x23x16 mm in size and hyperintense on T2-weighted sequences on the neck magnetic resonance imaging (MRI) (Figure 1). In the first evaluation of the MRI imaging, it was thought to be lymphangioma or hemangioma because of hypervascularisation but the MRI images were re-examined by experienced radiologists and it was decided to be a metastatic lymph node.

The fine needle aspiration (FNA) cytology of this lesion was positive for metastases of papillary carcinoma of the thyroid. Thus, thyroglossal duct cyst excision and bilateral total thyroidectomy with central and left neck lymph node dissection was performed.

Thyroglossal duct cyst specimen pathology has been reported as an invasion of the bone and soft tissue in the form of papillary thyroid carcinoma. All of the central lymph nodes were reactive and thyroid gland was normal but there was a metastatic lymph node in the lateral neck dissection which is 7 cm in diameter. After the operation, there was not a significant residual thyroid tissue and suspicious cervical lymph node by ultrasound imaging of the neck. One hundred and fifty mCi I-131 radioactive iodine ablation (RAI) therapy was given after the operation. Thyroid stimulating hormone (TSH) was over 100 mUI/L, but no evidence of metastasis was found.

One hundred and fifty mcg levothyroxine therapy was initiated after the RAI ablation and the patient was monitored under close follow-up.

DISCUSSION

TGDCs are the most common form of congenital cysts on the neck. These are the cysts of epithelial remnants of the thyroglossal tract and present characteristically as a midline neck mass, settled 61% in thyrohyoid region, 24% in suprahypothyroid region, 13% intralingual and 2% in the suprasternal region (1). TGDCs are usually within 2 cm of the midline (usually in the left) and are often associated with the hyoid bone, thus move up with swallowing or protrusion of the tongue. TGDCs are painless unless infected. Although most patients with TGDC are children or adolescents, up to one-third are aged 20 years or older. The wall of a TGDC is the second most common site for ectopic thyroid tissue (the first one is lingual thyroid).

The differential diagnosis of TGDCs are dermoid and sebaceous cysts, branchial cleft cysts which are placed in the midline, lipomas, hypertrophic pyramidal lobe and thyroid nodules, lymph nodes and salivary gland tumors located in the midline of the neck.

FNA is often used to diagnose TGDCs but it has moderate sensitivity. The cytomorphic features include colloid, macrophages, lymphocytes, neutrophils, and ciliated columnar cells, but these are not specific to TGDCs. Cytology is useful to exclude other diagnoses with an appropriate clinicoradiologic examination (2). USG is a very useful modality for thyroid gland and reveals the cystic nature of a TGDC, but does not provide information about the relationship to surrounding structures including hyoid bone, thus contrast enhanced computed tomography (CT) or MRI are more valuable than USG to determine the relationship with TGDC and other structures in the neck (3).

The standard operation is resection of the cyst and the midportion of the hyoid bone in continuity and resection of a core of tissue from the hyoid upwards towards the foramen cecum, an operation known as the Sistrunk procedure. When lymph node metastases are detected preoperatively, total thyroidectomy and regional neck dissection are recommended in addition to Sistrunk’s procedure. Suppressive hormone therapy and radioactive iodine are recommended in all post-thyroidectomy patients (4).

We performed Sistrunk procedure and total thyroidectomy with central and lateral lymph node dissection due to the presence of lymph node metastasis. Any metastatic lymph node was found in central compartment but there was metastasis in lateral compartment. After the operation RAI ablation therapy and then suppressive hormone therapy was given as recommended.

The incidence of primary carcinoma of the TGDC is less than 1% in all age groups (5,6,7). Eighty-eight percent is papillary carcinoma, 6% is squamous cell carcinoma, and the rest are Hurte cell, follicular and anaplastic carcinomas. Medullary thyroid carcinoma have not been reported in TGDCs (8).

In a series of 18 patients with papillary cancer arising in a TGDC, 16 patients underwent neck dissection of the central and/or lateral compartments. Tumor foci were found in the thyroid lobes in 9 of 16 patients (56%). Lymph node metastases were found in 12 of 16 (75%). Nodes were positive in 6 of 15 central compartment dissections (40%) and in 9 of 15 lateral neck dissections (60%). Metastases to the lateral compartment, with no central compartment metastasis, were found in 6 of 15 patients (40%) (9). Occurrence of distant metastasis is uncommon (5).

Our case is a rare case due to have a thyroid papillary carcinoma in a TGDC with lateral lymph node metastasis and without a tumor in thyroid gland and without central lymph node metastasis.

As a result, while evaluating a thyroglossal duct cyst, in all cases, a detailed neck ultrasonography should be performed and biopsy should be done from suspected lymph nodes before planning the surgery.

8- Thyroglossal duct cysts and ectopic thyroid. Sep 2012 UpToDate.