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Papillary Carcinoma of the Thyroglossal Duct Cyst: Case Report

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Abstract
Thyroglossal duct cysts 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%. In most cases the diagnosis is made postoperatively. We present a 22-year-old female with thyroid papillary carcinoma arising from thyroglossal duct cyst, identified in pathologic study after sistrunk operation. The patient then underwent total thyroidectomy and bilateral neck dissection. The patient was treated with radioactive iodide and thyroid suppression therapy was given as adjuvant treatment. The patient has been following for two years without any metastasis.

Keywords: Thyroglossal duct cyst, Thyroid papillary carcinoma, Thyroidectomy

Introduction
As the thyroid gland descends from the foramen cecum to its location at the point below the thyroid cartilage, it leaves behind an epithelial trace known as the thyroglossal tract. The tract disappears during the 5th-10th gestational week. Incomplete atrophy of the thyroglossal tract, or retained epithelial cyst, creates the basis for the origin of a thyroglossal duct cyst (1). A thyroglossal duct cyst is the most common anomaly in the development of the thyroid gland (2). Seventy percent are diagnosed in childhood and 7% are diagnosed in adulthood (3). Only 1% of thyroid carcinomas arise from a thyroglossal duct cyst (4).

Case Report
A 22-year-old female patient was admitted with the complaint of swelling in his neck. There was no history of dysphagia, hoarseness, or fever. In the physical examination, there was a painless cystic mass about 3.0 × 1.5 cm in size at midcervical line which moves with gulping. There was no other finding on the neck examination regarding lymphadenopathy and thyroid nodule. Routine blood investigations and thyroid function tests were normal. Ultrasonography of the neck showed a well-defined, cystic mass (23 × 18 × 12 mm in size) with an echogenic component (7 × 7 mm in size) (Fig. 1). The patient underwent sistrunk operation. The pathologic report showed duct with stratified squamous epithelium, mild infiltration of lymphocytes, fibrotic stroma and many true papillary structures that lined by cuboidal cells, indicating a malignant papillary carcinoma arising in a thyroglossal duct cyst (Fig. 2).
Total thyroidectomy and bilateral neck dissection were performed. The final postoperative pathology reported papillary carcinoma without any metastasis to the thyroid and cervical lymph nodes. One hundred mCi I-131 radioactive iodide and thyroid suppression therapy was given after the operation. The patient has been following without any metastasis for two years.

Fig. 1: Ultrasonography of the neck showed a cystic mass.

Fig. 2: Showing papillary carcinoma arising from a thyroglossal duct cyst.

Discussion

Thyroglossal duct cyst carcinoma typically presents as a midline palpable neck mass. Malignancy should be suspected if the cyst is hard, fixed, irregular, or displays sudden expansion with palpable neck lymph nodes (5). Malignant transformation is a rare complication of untreated thyroglossal duct cyst.

The diagnosis is, however, often made postoperatively on histologic examination of the resected specimen. In one study the age at presentation ranged from 6 to 81 years, with an average of 39 years. Women are affected more often than men (6). To confirm a diagnosis of thyroglossal duct cyst, the following criteria should be fulfilled: the cyst must be located in the median region of the neck; the cyst wall must be composed of cuboidal epithelial cells; and lymphatic tissues and normal thyroid follicles must be present in the cystic wall (7). Malignant tumors developing from the thyroglossal duct have two origins: thyrogenic carcinoma arising from thyroembrionic remnants in the duct or a cyst, and squamous cell carcinoma arising from metaplastic columnar cells that line the duct (1). More than 200 cases of thyroglossal duct carcinomas have been reported in which papillary carcinoma accounts for 80% of cases, with the rest being squamous cell carcinoma (8).

In our case, the presence of carcinoma within a thyroglossal duct and normal thyroid tissue support the diagnosis of papillary carcinoma arising in a thyroglossal duct remnant rather than a metastasis from a primary carcinoma of the thyroid gland. On ultrasonography, a benign thyroglossal duct cyst can be anechoic, homogenously hypoechoic, homogenously hyperechoic, or heterogenous in appearance. If calcification is seen within, malignancy needs to be suspected. Calcification is the hallmark of papillary carcinoma in a thyroglossal duct cyst (9). In our case, apart from the cystic lesion, ultrasonography showed an echogenous component without calcification. The common surgical procedure used for a thyroglossal duct cyst is sistrunk’s procedure. In case of malignancy, additional steps should consist of thyroidectomy, radioactive iodine and thyroid suppression (1). In our case, this patient had no nodules in the thyroid gland, no previous exposure to radiation, no lymph node metastasis and no extra capsular spread of the thyroglossal duct cyst carcinoma.

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She needs further life-long follow-up with physical examinations and imaging studies.

Ethical considerations

Ethical issues (Including plagiarism, Informed Consent, misconduct, data fabrication and/or falsification, double publication and/or submission, redundancy, etc) have been completely observed by the authors.

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The authors declare that there is no conflict of interests.

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