CASE REPORT
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A Case of Thyroglossal Duct Papillary Carcinoma with Subsequent Lateral Neck Node Metastases

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Thyroglossal duct carcinoma is very rare, being present in about 1% of all patients with thyroglossal duct cysts. The clinical presentation of thyroglossal duct carcinoma is very similar to that of its benign counterpart; thus, a diagnosis of malignancy is usually made on postoperative histopathological examination. In general, the Sistrunk operation is sufficient for patients with no signs of invasion. However, total thyroidectomy and appropriate neck dissection are required if there are intra-thyroidal lesions or evidence of cervical lymph node metastasis. We experienced a case of 50-year-old female patient with a papillary carcinoma arising in the thyroglossal duct cyst and subsequent metastases to lateral neck nodes.

Key Words: Thyroglossal duct cyst, Papillary carcinoma, Sistrunk operation

Introduction

Although thyroglossal duct cyst is the most common congenital lesion in the cervical area, constituting an estimated 75% of cervical masses, primary carcinoma arising in the thyroglossal duct cyst is very rare, presenting in less than 1% of thyroglossal duct cysts.[1-5]

Since its first description in 1911,[6] approximately 240 cases of thyroglossal duct carcinoma have been reported in the English literature: We present a rare case of papillary carcinoma arising in a thyroglossal duct cyst with subsequent lateral neck node metastases. Written informed consent was obtained from the patient for publication of this case report.

Case Report

A 50-year-old female patient presented with a palpable cystic mass in the submental region for 3 months. This patient had no conventional symptoms, and no specific medical history.

Cervical ultrasonography showed a 4.0 cm sized, well-demarcated, cystic mass and a small mural lesion on the submental area, but fine needle aspiration cytology revealed no evidence of malignancy. She was diagnosed as having a thyroglossal duct cyst, and a Sistrunk operation was performed. Operative findings demonstrated a 4.0 cm sized cystic mass containing dark green fluid and passing through the hyoid bone. The mid-portion of the hyoid bone was excised along with the cyst, and the cyst was traced and ligated near the foramen cecum.

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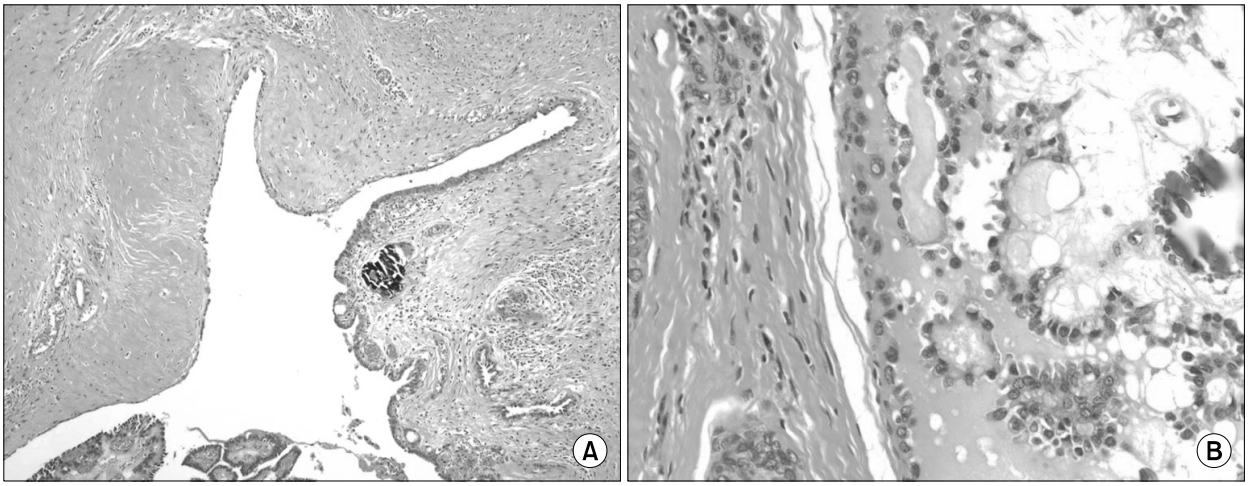

Fig. 1. Histologic findings of the thyroglossal duct carcinoma. (A) Other part of the thyroglossal duct reveals cystic structure with papillary carcinoma (H&E, ×100), and (B) High power view of the papillary carcinoma show a typical papillary structure and psammoma body (H&E, ×400).

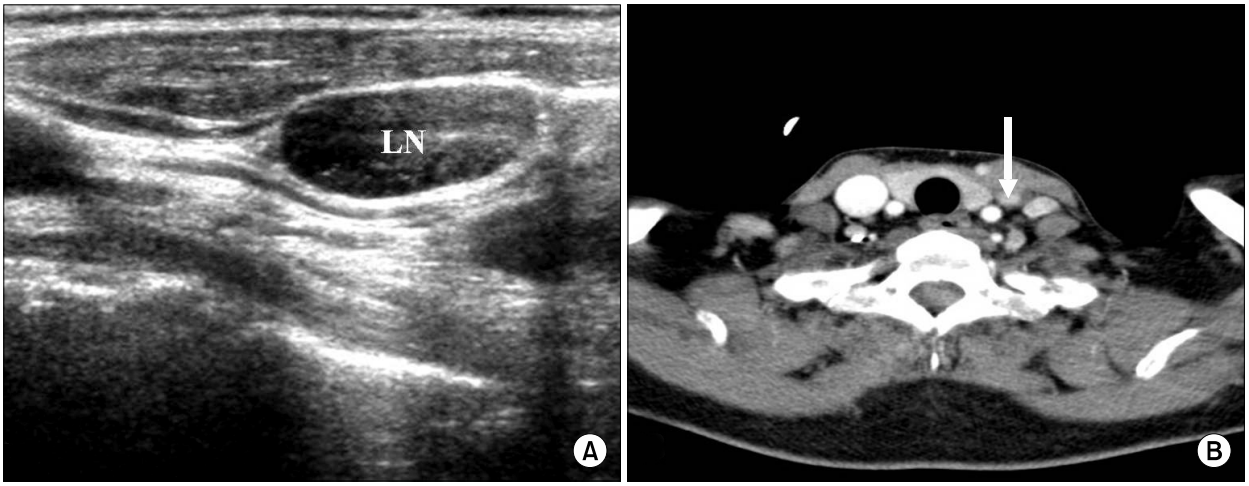

Fig. 2. Cervical ultrasonography (A) and computed tomography (B) demonstrating the enlargement of left lateral lymph nodes (white arrow).

Histologically, the tumor was diagnosed as a thyroglossal duct cyst with histologic evidence of papillary carcinoma arising from the thyroglossal duct cystic wall (Fig. 1).

Four years after the initial operation, an enlarged lymph node at the level III on the left neck was seen on follow-up neck ultrasonography and computed tomography (Fig. 2). Fine-needle aspiration cytology suggested metastatic papillary thyroid carcinoma.

Thus, a total thyroidectomy along with left modified neck node dissection was performed. Histological examination revealed no malignant lesions in the thyroid gland, but three metastatic papillary carcinomas were found in the left level III lymph nodes (3/20) (Fig. 3).

Two months later, she underwent radioactive iodine ablation, and the iodine-131 whole body scan revealed no abnormal uptake in the thyroid bed or other areas. The patient remains free of disease three years after the second surgery.

Discussion

Thyroglossal duct cysts are the most common congenital anomalies of neck, accounting for 75% of

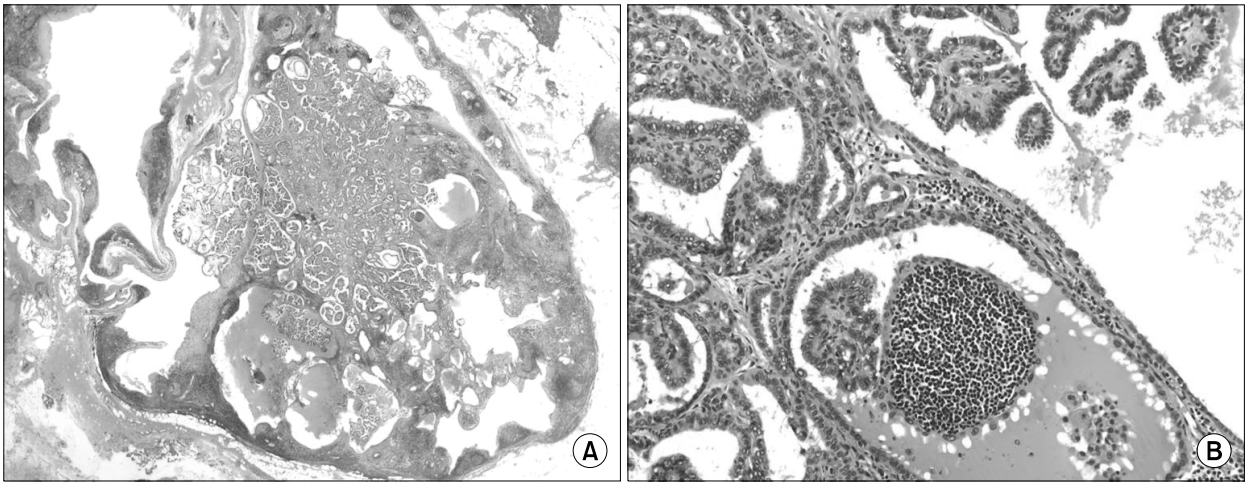

Fig. 3. Histologic findings of the metastatic lymph node. (A) The cystic papillary carcinoma occupies the entire lymph node (H&E, ×12.5), and (B) High power of the papillary carcinoma show a typical papillary structure and cytologic characteristics (H&E, ×400).

congenital cervical masses.[1,4] However, carcinoma arising in a thyroglossal duct cyst is extremely rare, occurring in less than 1% of thyroglossal duct cysts in adults.[2,3]

The median age for development of carcinoma in the thyroglossal duct cyst is during the fourth decade of life, and these tumors have a slight female predominance (female : male=3 : 2).[4,7,8]

Thyroglossal duct carcinoma is therefore generally of thyroid or squamous cell origin. Papillary carcinoma is the most common type (80%), followed by mixed papillary/follicular carcinoma (8%) and squamous cell carcinoma (6%). The other 6% include Hurthle cell, follicular, and anaplastic carcinoma.[8,9]

Thyroglossal duct carcinoma is usually diagnosed by an incidental finding after surgical excision, because the fine-needle aspiration cytology is useful in not helpful on a preoperative diagnosis.[2]

The criteria that should be fulfilled for a definitive diagnosis of primary thyroglossal duct carcinoma include; (1) location in the wall of the thyroglossal duct cyst; (2) differentiation from a cystic lymph node metastasis by histologic demonstration of a squamous or columnar epithelial lining and normal thyroid follicles in the wall of the cyst; and (3) no malignancy in the thyroid gland or any other possible primary site.[10] Synchronous carcinoma in a thyroglossal duct cyst and thyroid gland has been reported in 33~40% of cases, cervical lymph node metastasis in 16~25% of cases and distant metastases in 1.3% of cases.[2,11,12]

The debate concerning the optimal management of thyroglossal duct carcinoma still exists. The first step for treatment of a thyroglossal duct carcinoma is the Sistrunk prodecure excision of the thyroglossal duct cyst and the midportion of the hyoid bone through or near which it traverses. It has been reported that the Sistrunk procedure is sufficient for patients with thyroglossal duct carcinoma with a good outcome.[7]

The crucial problem is the management of the thyroid gland. Total thyroidectomy enables the use of postoperative radioactive iodine treatment and a good follow-up with radioiodine scans and serum thyroglobulin concentrations.[2,11]

Some authors recently recommend a treatment strategy based on risk stratification in which high-risk patients undergo more aggressive treatment.[3,4] If any of the followings are present, total thyroidectomy should be recommended: (1) patient older than 45 years; (2) history of radiation exposure; (3) clinically or radiologically evident thyroid lesions; (4) suspicious lymph node metastasis; (5) tumor more than 1.5 cm in diameter; and (6) cyst wall invasion and positive margins on postoperative histologic examination.[4]

As for lymph node metastases, routine selective neck dissection is not recommended. Selective neck dissection should be performed only in cases of

clinically evident lateral lymph node metastases.[3,7,13]

Postoperative radioactive iodine treatment should be given to patients undergoing total thyroidectomy. In the present case, the patient underwent the Sistrunk procedure initially because there was no evidence of thyroid lesions. Total thyroidectomy plus lateral neck dissection was performed four years later because of the lateral node metastasis from thyroid carcinoma.

The prognosis for patients with thyroglossal duct carcinoma is excellent, with an overall survival rate of 100% at 5-year and 95.6% at 10-year,[7] and disease-related mortality has been reported only in few cases.[13]

In conclusion, although malignancy occurring in the thyroglossal duct cyst is extremely rare, patients can be correctly managed by maximizing the awareness of this entity, careful clinical examination and imaging when appropriate, as well as proper surgical treatment.

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