

Ectopic Thyroid Papillary Carcinoma Presenting as Bilateral Neck Masses

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Ectopic thyroid papillary carcinoma presenting as bilateral neck lymph nodes metastasis is very rare. We report a 42-year-old male with midline submental ectopic thyroid papillary carcinoma presenting as bilateral progressively growing neck masses for 3 months. A right neck mass excisional biopsy was carried out and the pathology revealed metastatic papillary carcinoma. Total thyroidectomy, submental “ectopic thyroid tissue” excision and bilateral neck dissection were then performed. No primary lesions were found in the thyroid gland. Therefore, the differential diagnosis of bilateral neck masses should include cervical metastasis of ectopic papillary carcinoma. [*J Chin Med Assoc* 2010;73(4):219–221]

Key Words: cervical lymph node metastasis, ectopic thyroid, head and neck, papillary thyroid carcinoma, thyroid cancer

Introduction

Ectopic thyroid tissue may appear in any location along the trajectory of the thyroglossal duct from the foramen cecum to the mediastinum, and a thyroglossal duct cyst is the most common congenital anomaly. Ectopic thyroid carcinoma is rare, but commonly presents as a midline cervical mass.¹ Among ectopic thyroid carcinomas, papillary carcinoma comprises the majority of cases. Although papillary carcinoma of the thyroid gland has a tendency toward neck metastasis, ectopic thyroid papillary carcinoma with cervical lymph node metastasis as the initial presentation is extremely rare. We describe a rare case of ectopic thyroid papillary carcinoma presenting as bilateral cervical metastases.

Case Report

A 42-year-old male presented with a slowly growing right neck mass for 3 months. He had a history of goiter with regular follow-up at another hospital for 4 years, including negative findings of sonoguided fine needle aspiration several times. There was no history of radiation exposure. The patient was then referred to our clinic. Physical examination revealed bilateral cervical

masses at bilateral level III, 2 × 3 cm on the right and 1 × 1 cm on the left, with a rubber-like consistency. Nasopharyngoscopy revealed negative findings and thyroid function was normal. Neck computed tomography showed a submental 1.6 cm round, well-enhanced mass (Figure 1A) and bilateral neck levels III to IV multiple neck masses (Figure 1B).

Lymphoma was suspected and right neck mass excision was carried out. Three lymph nodes were removed, and 2 of them showed metastatic papillary carcinoma of the thyroid. Among the positive lymph nodes, 1 also showed focal extracapsular spread. Thyroid sonography was performed to determine the location of the primary tumor in the thyroid gland, and bilateral small hypoechoic nodules were observed. Therefore, another operation including total thyroidectomy, bilateral selective neck dissection (levels II–IV and VI), and submental “ectopic thyroid tissue” excision was carried out. The left level III neck mass showed 3 positive lymph nodes for metastatic papillary carcinoma among a total of 6 lymph nodes, but there was no primary papillary carcinoma in the thyroid gland. However, 4 foci of multicentric papillary carcinoma among the normal thyroid tissue were found in the submental mass and measured 1.2 × 1.4 cm without thyroid capsular invasion, although there was no visible



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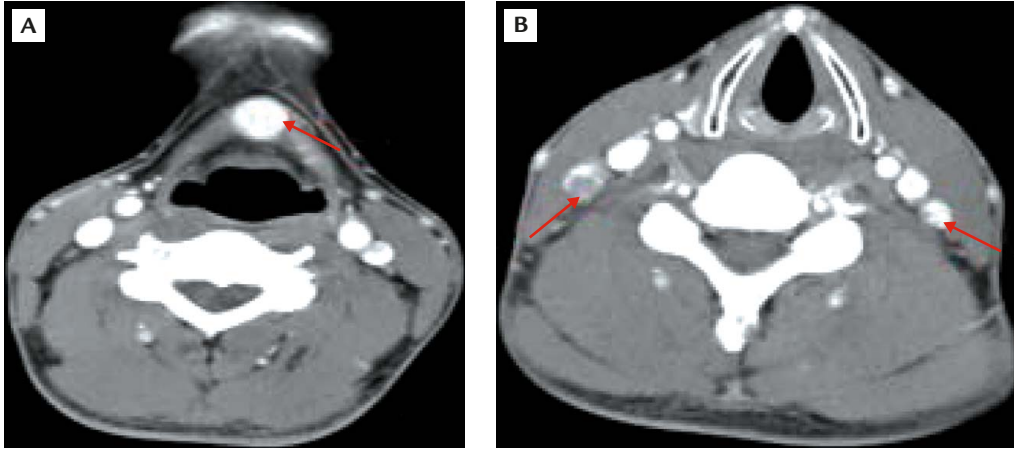


Figure 1. Neck contrast computed tomography of a 42-year-old male who initially presented with bilateral cervical masses shows a submental mass. (A) A 1.6-cm well-enhanced nodule over the submental area (arrow). (B) Bilateral levels III-IV lymphadenopathies (arrows).

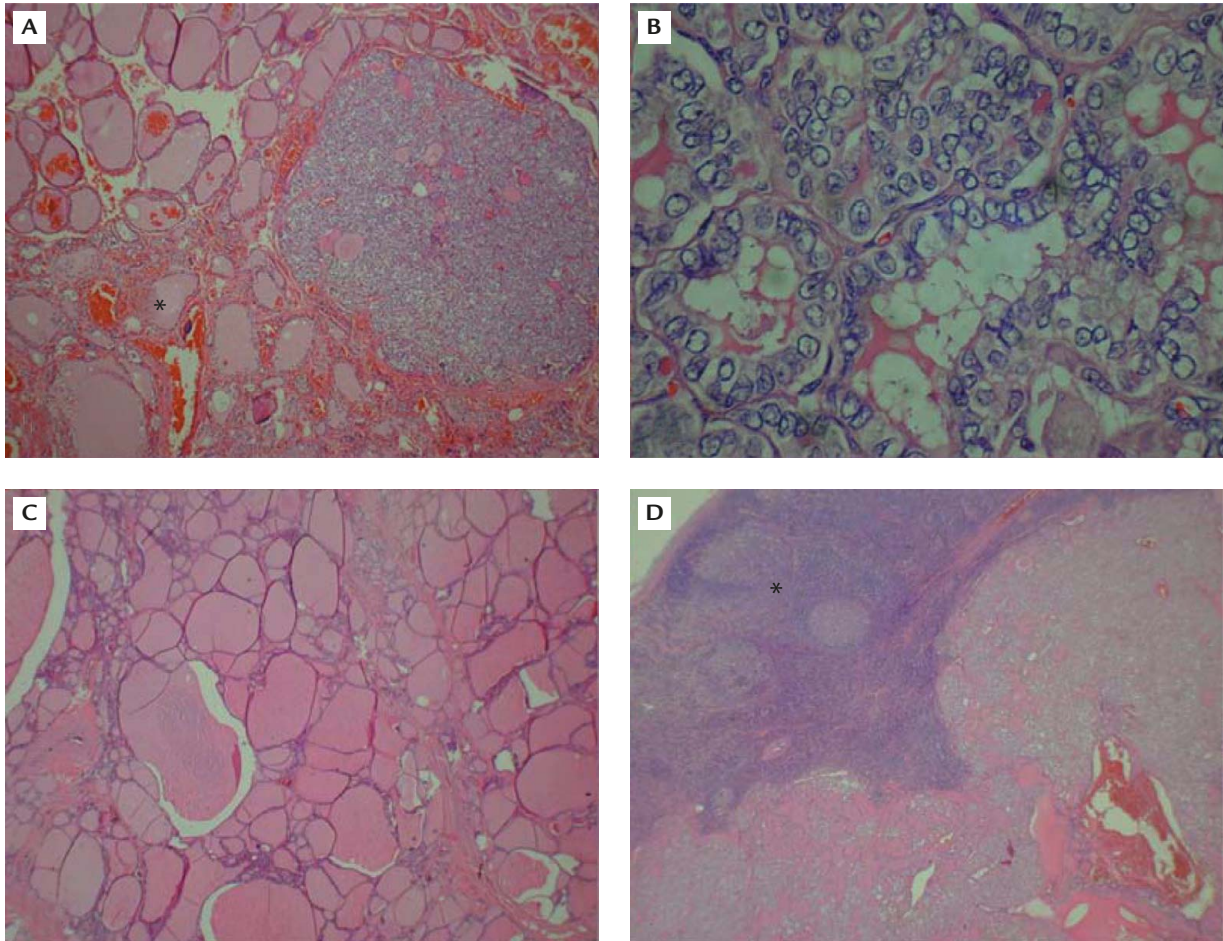


Figure 2. (A) Ectopic thyroid papillary carcinoma with surrounding normal thyroid tissue (asterisk) [hematoxylin & eosin (H&E), 100×]. (B) Ectopic thyroid papillary carcinoma (H&E, 200×). (C) Thyroid gland: nodular goiter (H&E, 100×). (D) Metastatic papillary carcinoma in a cervical lymph node (asterisk) (H&E, 40×).

thyroglossal duct cyst or lymph node structures (Figure 2). Therefore, we revised the diagnosis as ectopic thyroid papillary carcinoma with bilateral cervical lymph node metastases.

I-131 ablation (100 mCi) was carried out 6 weeks later and a post-ablation scan showed uptake over the anterior lower neck and upper mediastinum. A thyroxine supplement was then prescribed. Two follow-up

I-131 whole-body scans were performed at 14 and 26 months later, and both showed no further uptake. Serum thyroglobulin levels under thyroxine supplement were also within the normal range.

Discussion

In a clinical evaluation of a recent onset of bilateral neck masses among older adults (≥ 40 years old), malignancy should be considered first, including lymphoma and metastatic carcinoma. Other etiologies include infectious lymphadenitis (granulomatous lymphadenitis) or, rarely, bilateral carotid body tumors.² In a review study, 163 community-based patients who had asymmetric neck masses underwent neck biopsies, and the outcomes showed 29.4% metastatic carcinomas and 21.4% lymphomas.³ These findings are consistent with the 50% incidence rate of malignancy in adult patients.

In the present case, a neck mass excisional biopsy was carried out directly because lymphoma was initially suspected due to the rubber-like consistency of the bilateral cervical masses. However, metastatic papillary carcinoma was indicated by the pathology. Cervical lymph node involvement at presentation has been reported to be approximately 30% of thyroid papillary carcinoma cases.⁴ In some cases of cervical metastatic papillary carcinoma, no detectable primary lesion in the thyroid gland can be found by ultrasonography. Occult papillary carcinoma has been defined for such cases in the literature,⁵ and our patient initially fit the definition. It has been reported that primary thyroid papillary carcinoma is found in 64% of cases of occult papillary carcinoma after total thyroidectomy.⁵ However, we did not find any primary thyroid lesion after total thyroidectomy, even though a detailed pathology review was made. Surprisingly, primary papillary carcinoma was found in the submental ectopic thyroid tissue.

Ectopic thyroid tissue can be found anywhere from the tongue base to the mediastinum, and the most common type is a thyroglossal duct cyst. The incidence of carcinoma is less than 1% in thyroglossal duct cysts.⁶ The most common histologic subtype is papillary carcinoma, occurring in over 90% of thyroglossal duct cyst carcinomas, including the mixed forms.⁷ Most thyroglossal duct cyst carcinomas are located in the thyrohyoid region, similar to this case. Despite a comprehensive pathology review in this case, the submental mass showed both the structure of papillary carcinoma

and normal thyroid tissue, without a thyroglossal duct cyst or lymph node features. Suda et al have also reported a case of submental ectopic thyroid papillary carcinoma lacking a thyroglossal duct cyst but without bilateral neck lymph nodes as the first presentation.⁸

The incidence of cervical lymph node metastases from ectopic thyroid papillary carcinoma has been estimated to be approximately 8%.^{7,9-11} Ectopic thyroid papillary carcinoma presenting as bilateral neck metastasis, as in our case, is even rarer. In the treatment of papillary carcinoma with neck metastasis, lateral neck dissection should be included with total thyroidectomy, bilateral central compartment dissection, and post-operative radioactive iodine ablation therapy.¹² There was no evidence of locoregional or distant metastasis 26 months after 1 course of I-131 ablation therapy. In summary, we have presented a rare case of ectopic thyroid papillary carcinoma presenting as bilateral cervical lymph node masses. The differential diagnosis of bilateral neck masses should include metastasis from ectopic thyroid papillary carcinoma.

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