Case Report



# Occult Papillary Thyroid Carcinoma Presenting as Huge Cervical Metastasis with Cystic Pattern

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## Article info

## Article history:

Received: September 10, 2007 Revised: September 28, 2007 Accepted: November 14, 2007

*Keywords:* Cervical metastasis Cyst Cystic metastasis Papillary carcinoma Thyroid carcinoma

## Abstract

Cervical cysts are usually benign in the population younger than 40 years. However, neck metastasis from malignancies of the head and neck may present as cervical cysts. Here, we report a case of papillary thyroid carcinoma with cervical metastasis and cystic change. A 35-year-old man visited our clinic, and his chief complaint was related to the presence of a huge neck mass for 15 years. Computed tomography scans revealed cystic masses and abnormal calcification in the thyroid gland. Fine needle aspiration cytology of the left thyroid gland revealed atypical cells with a few cells harboring intranuclear pseudoinclusion. We then performed dissection of the left side of the neck to remove the cystic masses and a left thyroid lobectomy. Intraoperative frozen section biopsy of the cystic masses showed benign cystic lesions. However, the results of the final pathologic examination proved thyroid papillary carcinoma in the thyroid gland and the cystic masses. The patient received dissection of the right side of the neck accompanied by right thyroid lobectomy. After the operation, <sup>131</sup>I ablation therapy was performed. There was no recurrence noted during the regular 2-year follow-up period. (Tzu Chi Med J 2008;20(2):140-143)

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# 1. Introduction

Cystic lesions of the neck in the young adult population (16–40 years) are usually benign. Cystic lesions of the neck have rarely been proven to metastasize from squamous cell carcinoma of the oronasopharyngeal area, or from papillary thyroid carcinoma (1,2). Cystic neck masses in a young adult patient may pose a diagnostic challenge because the primary sites of malignancy may not be detected using routine physical examination and fine needle aspiration (FNA) often fails to give a definitive diagnosis (3). The medical history might also be misunderstood. Excision biopsies are required to confirm the final diagnosis. Therefore, we present a case of long-standing huge cervical cysts as the initial manifestation of papillary thyroid carcinoma.

## 2. Case report

A 35-year-old man presented to our department in March 2003 for evaluation of bilateral neck masses.



Fig. 1 — Axial computed tomography section with contrast shows well-defined cystic masses (arrows) in the left neck and high-density material attached to the cyst wall (arrowhead).



Fig. 3 -Gross view of the cystic masses after left neck dissection.



Fig. 2 — Axial computed tomography section with contrast shows calcification in the left lobe of the thyroid gland (arrow).

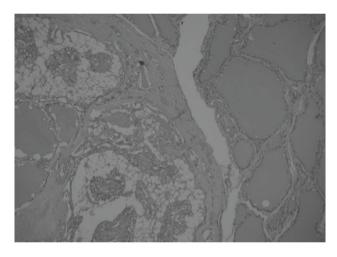


Fig. 4 — Papillary thyroid carcinoma composed of complex, branching and randomly oriented papillae with fibrovascular cores.

He had noticed his neck masses for 15 years but he did not seek any medical attention. There were no associated symptoms. Clinical examination results showed two masses, one measuring  $10.0 \times 6.0$  cm and the other  $4.0 \times 4.0$  cm over the left neck, and a  $3.0 \times 3.0$  cm mass in the level III area over the right neck. The masses were smooth, nontender and soft. Physical examination results of the thyroid gland and fiberscopic examination of the oronasopharyngeal area were essentially normal. Computed tomography (CT) scans of the neck revealed oval-shaped cystic masses underneath the left sternocleidomastoid muscle and high-density contents attached to the cyst wall (Fig. 1). There was also abnormal calcification in the left lobe of

the thyroid gland (Fig. 2). FNA cytology of the thyroid gland revealed atypical cells with a few cells harboring intranuclear pseudoinclusion.

Under the impression of cystic hygroma of the neck and thyroid tumor, we explored the neck surgically. Frozen section biopsy of the neck mass showed benign cystic lesions, and the neck masses were excised with walls intact (Fig. 3). Thyroid lobectomy on the left side was also carried out and firm nodules in the left lobe of the thyroid gland were found during the operation. The final pathologic results revealed papillary thyroid carcinoma measuring  $4.0 \times 3.6 \times 2.2$  cm inside the thyroid tissue, and metastatic papillary carcinoma in the neck masses (Figs. 4 and 5). Thereafter, we carried

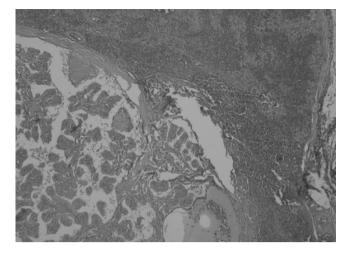


Fig. 5 — Metastatic papillary carcinoma in neck lymph nodes.

out total thyroidectomy with bilateral modified neck dissection. The results of the right thyroid lobe and the dissected lymph nodes also showed papillary thyroid carcinoma and metastatic carcinoma.

After surgery, the patient received <sup>131</sup>I (90mCi) ablative therapy. <sup>131</sup>I whole body bone scan revealed no metastasis. We continued to follow the patient and no recurrence or morbidity was noted after surgery. Finally, follow-up serum thyroglobulin level was 100 ng/ mL under thyroxine treatment.

## 3. Discussion

Papillary carcinoma is the most common type of thyroid malignancy, usually presenting as a thyroid nodular lesion. It may present as a mass over the neck without the typical pattern of a thyroid neoplasm (4–7). Incidence of cervical lymph node involvement with papillary thyroid carcinoma has been reported to be 44-80% (8). Such metastatic lymph nodes usually appear as solid masses in the anterior or lateral aspect of the neck. However, the metastatic lymph nodes could undergo cystic degeneration and present as cystic masses in the lateral aspect of the neck. Fewer than 35 patients with the above presentation have been reported in the literature (1). In addition, thyroid papillary carcinomas are generally slow-growing neoplasms. Long-standing cervical cystic metastases may give physicians the wrong impression of benign cervical cysts and could possibly cause a delay in diagnosis.

Lateral cervical cysts are often considered to be benign lesions in young people (16–40 years) (2). Cervical cysts in 90% of patients in the young adult population were proven to be benign, and the most common diagnosis was branchial cleft cyst (1). It is well known that carcinoma that metastasizes to lymph nodes may undergo cystic degeneration. This condition is less frequent and might be caused by primary branchiognic carcinoma, or metastatic tumors arising mainly from carcinomas in Waldeyer's ring (tongue base, tonsil, nasopharynx) and the thyroid and salivary glands (2,3). In particular, Seven et al reported an incidence of 11% (4/37) of thyroid malignancy in those with cervical cysts (1). The primary sites of malignancy may not be detected by careful inspection and palpation, especially in small primary tumors of the thyroid gland. Therefore, it is not very easy to differentiate benign cervical cysts from cervical metastases of malignancies based on history or physical findings. Thus, several authors advocate that cervical cysts should be presumed to be carcinoma until proven otherwise, even after thorough head and neck examination results reveal negative findings for primary neoplasms (3,9).

FNA is a useful diagnostic tool for neck masses, with reported sensitivity rates ranging from 90% to 100% (1). In solid lesions of the neck, the specificity is 100%, and the false-negative rate is <3% (3). FNA is less useful in the diagnosis of cystic lesions, with a false-negative rate of 50-67% (3). Therefore, a negative report on FNA cytology in cystic lesions must be considered as inconclusive. In the presented case, we did not check the preoperative thyroglobulin level in cystic fluid. However, recent reports in the literature have shown that thyroglobulin from FNA biopsy was an effective method to diagnose metastatic cervical lymph nodes from papillary thyroid carcinoma (10). Cignarelli et al concluded that high thyroglobulin values from FNA biopsies with nondiagnostic cystic cytology strongly suggested cystic metastatic papillary thyroid carcinoma (10).

CT is one of the most important tools for the investigation of neck masses and cysts. Branchial cleft cysts are characteristically well-defined, low-density unilocular masses with thin uniform enhancing rims on CT scans. Wunderbaldinger et al reported that in young patients, cystic metastasis of papillary thyroid cancer might appear purely cystic, thereby mimicking branchial cysts (11). Two recent reports suggested that atypical findings, such as the presence of intracystic solid components or thickened or irregular outer walls on CT, should raise the suggestion of malignancy (1,12). In the present case report, a CT scan was obtained preoperatively and revealed cystic masses with thick and irregular margins with clearly intracystic enhanced elements. The above CT findings are compatible with those reported in previous studies. Therefore, atypical findings such as the presence of thickened or irregular cystic walls or intracystic enhanced elements on CT scans may indicate metastases from a thyroid malignancy.

Diagnostic approaches and management of cervical cysts are described in the following paragraph to

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help prevent inadvertent removal of metastatic lesions. First, a thorough head and neck examination should be performed to identify the primary carcinoma. Imaging studies such as CT/MRI should be carried out to identify the primary site and to evaluate the cystic mass (13). FNA of the mass for cytology may be helpful, but a negative report must be considered inconclusive (9). If the FNA is negative for malignancy, a frozen section biopsy of the cystic mass in the operating room should be performed. If the frozen section biopsy is positive for metastatic squamous cell carcinoma, a neck dissection should be performed after a panendoscopy of the head and neck and biopsy of suspicious sites (13). If the frozen section biopsy proves to be metastatic papillary thyroid carcinoma, total thyroidectomy and neck dissection should be performed (7,8). However, it is controversial as to whether or not frozen section biopsy should be routinely performed for cystic masses in the young adult population (16-40 years) because 90% of the cysts are benign (3). This approach should be used if suggestion of malignancy is prominent (1).

In conclusion, primary thyroid carcinoma with neck metastasis should be considered in a patient with a long history of huge cervical cysts until proven otherwise. A complete search for the primary site must be done and should include the thyroid gland. Preoperative FNA cytology and frozen section biopsies during surgery are helpful; however, negative results should be considered inconclusive. Thyroglobulin from FNA biopsy is reported to be helpful, and could be a fast procedure to diagnose metastatic cervical lymph nodes from papillary thyroid carcinoma. Care must be taken in the evaluation of patients with suggested carcinoma because inadvertent removal of neck metastasis or a delay in diagnosis will negatively affect prognosis.

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