1 Submandibular solid-cystic mass as the first and sole manifestation of occult thyroid papillary carcinoma: A case report

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Abstract

BACKGROUND

Occult thyroid papillary carcinoma (TPC) is always characterized by cervical lymph node metastasis as the initial manifestation, and TPC can be found by ultrasound. However, submandibular solid-cystic mass was the initial and sole manifestation, high frequency ultrasound, enhanced multislice CT scan and thyroid function tests showed no abnormalities in the thyroid gland, which is relatively rare.

CASE SUMMARY

A 24-year-old Chines female, who studied at university in Shandong Province, presented to clinic in June 2019 with a right submandibular mass which she had noticed 2 mo earlier. Clinical examination revealed a 2-cm, nontender, movable solid-cystic mass in the submandibular region, without palpable thyroid mass was observed. Ultrasonography revealed a 2.0*1.1cm solid-cystic mass in the right submandibular region, and the thyroid gland had no abnormalities. CT scan and 131I-WBS showed that there were no abnormalities in the thyroid. However, Cytology and pathology showed papillary tumor cell clusters, consistent with papillary thyroid carcinoma. So we perform total thyroidectomy and right neck lymph node dissection, the pathology
revealed thyroid were detected as classical thyroid micropapillary carcinoma, and lymph nodes of the level of VI central and level II ,III,IV, V right group showed no tumor metastasis. The patient was followed up for 2 years without significant recurrence.

CONCLUSION
Submandibular solid-cystic mass as the first and sole manifestation of occult thyroid papillary carcinoma is relatively rare. For the neck masses fine needle aspiration is necessary.

Key Words: occult thyroid carcinoma, submandibular mass, ultrasound, fine needle aspiration, case report

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Core Tip: Occult thyroid papillary carcinoma (TPC) is always characterized by cervical lymph node metastasis as the initial manifestation, and TPC can be found by ultrasound. However, submandibular solid-cystic mass was the initial and sole manifestation, high frequency ultrasound, enhanced multislice CT scan and thyroid function tests showed no abnormalities in the thyroid gland, which is relatively rare. We report the case with submandibular solid-cystic mass as the first and sole manifestation was diagnosed with papillary thyroid carcinoma after surgery.

INTRODUCTION
Thyroid carcinoma is a prevalent endocrine malignancy in clinical practice, with papillary thyroid carcinoma (PTC) being the most common subtype, accounting for approximately 90% [1-2]. Despite the favorable prognosis of PTC, early lymph node metastasis occurs in 20-90% of cases [3-6]. Lymph node metastasis in PTC predominantly
involves multiple regions, while single-region metastasis is rare. Metastatic involvement commonly affects the central VI level and lateral groups II, III, and IV; however, it is infrequent in levels V and I\textsuperscript{17}. Occult thyroid papillary carcinoma (OTPC) refers to any papillary thyroid carcinoma with a diameter less than 1 cm. OTPC typically presents initially as cervical lymph node metastasis but rarely manifests as a submandibular solid-cystic mass. Palpation alone may not effectively detect OTPC. Most thyroid nodules or ectopic thyroids can be identified using high-frequency ultrasound imaging, enhanced multislice CT scanning or 131I whole-body follow-up scan (131I-WBS) \textsuperscript{[8-10]}, although some cases of OTPC remain undetected \textsuperscript{[11]}. Therefore, determining the necessity for regular follow-up and whether thyroid surgery should be performed are crucial topics along with defining the extent of lesion resection and cervical lymph node dissection scope. In this particular case study, an initial presentation solely consisting of a submandibular solid-cystic mass was observed without any abnormalities detected through high-frequency ultrasound imaging, enhanced multislice CT scanning or thyroid function tests—a relatively uncommon occurrence.

**CASE PRESENTATION**

**Chief complaints**
a right submandibular mass that had been noticed by her two months prior

**History of present illness**
a right submandibular mass had been noticed by her two months prior, ultrasonography demonstrated a 2.0*1.1cm solid-cystic mass in the right submandibular region while revealing no abnormalities in the thyroid gland (Figure 1). US-guided fine-needle aspiration (FNA) exhibited papillary tumor cell clusters consistent with papillary thyroid carcinoma.

**History of past illness**
There was no special history of past illness.

**Personal and family history**
There was no personal or family history of thyroid or other neoplasms.

**Physical examination**
Clinical examination revealed a 2-cm, non-tender, mobile solid-cystic mass in the right submandibular region without any palpable thyroid nodules.

**Laboratory examinations**
US-guided fine-needle aspiration (FNA) exhibited papillary tumor cell clusters consistent with papillary thyroid carcinoma.

**Imaging examinations**
Ultrasonography demonstrated a 2.0*1.1cm solid-cystic mass in the right submandibular region while revealing no abnormalities in the thyroid gland (Figure 1). $^{131}$I-WBS showed no abnormalities in the thyroid gland or any abnormal radioactive elevation in other parts of the body.

**FINAL DIAGNOSIS**
The pathology revealed metastatic carcinoma in lymphoid tissue with morphology consistent with thyroid papillary carcinoma metastasis (Figure 2).

**TREATMENT**
After adequate preoperative preparation, the patient opted for total thyroidectomy and right neck lymph node dissection. Postoperative radioiodine therapy and TSH inhibition therapy were administered after surgery.

**OUTCOME AND FOLLOW-UP**
Pathology identified a 0.3 cm nodule adjacent to the capsule as classical thyroid micropapillary carcinoma that invaded the capsule in the left lobe of the thyroid gland. Additionally, a 0.1 cm micropapillary carcinoma was found in the left lobe of the thyroid gland. Fortunately, cervical lymph nodes at level VI central and levels II, III, IV, V on right side showed no tumor metastasis but chronic inflammation. The patient has been followed up for 2 years without significant recurrence.

DISCUSSION

For the progressive enlargement of masses in the submandibular region, particularly in young patients, we recommend increased attention. Although most are benign lumps, such as submandibular lymphadenitis, submandibular cysts, and hypogland adenitis, there have been reported cases of malignant or metastatic lymph nodes. Ether reported a case where a right submandibular mass was diagnosed as mammary analogue secretory carcinoma through fine needle aspiration [12]. Bo-Jung reported two cases of nodal diffuse large B-cell lymphoma with morphologic features resembling primary mediastinal large B-cell lymphoma presenting in the submandibular lymph nodes [13]. Miles demonstrated that a right submandibular mass confirmed on histological analysis following excision of the right submandibular gland was multinodular adenomatous oncotic hyperplasia [14]. Performing high-frequency ultrasound for evaluating submandibular masses is necessary to differentiate between benign and malignant nodules. Sonographic features suggestive of malignancy or metastatic lymph nodes include enlargement, irregular borders, round shape, ill-defined contours, absence of an echogenic hilum, worsening microcalcification within lymph nodes, cystic areas within lymph nodes and color Doppler abnormalities such as hypervascularity [15-16]. If ultrasound fails to distinguish between benign and malignant lesions further cytology is recommended. In cases where ultrasound or cytology indicates metastasis excisional biopsy should be performed for definitive diagnosis. In our present case study, ultrasound revealed a solid-cystic mass with separate vascularity and clear border indicating potential malignancy. To establish a definitive
diagnosis US-guided fine-needle aspiration was performed which showed papillary tumor cell clusters consistent with papillary thyroid carcinoma. Further surgical intervention indicated lymph node metastasis from papillary thyroid carcinoma. Some studies have shown that PTCs frequently undergo cystic trans-formation, both in the primary tumor and in the metastatic lymph node [17], which is consistent with this case.

OTPC is relatively infrequent in clinical practice; it commonly presents with neck lymph node metastasis as an initial symptom but can occasionally manifest as lung or bone metastasis [18-19]. High-frequency ultrasound serves as the gold standard for diagnosing thyroid cancer by enabling detection of lesions as small as 3mm [20]. Thyroid cancer exhibits irregular margins, hypoechoigenicity, and microcalcifications. Malignant nodules demonstrate significantly greater hardness than benign ones. Real-time shear wave elastography (SWE) plays a crucial role in differentiating between benign and malignant nodules [21]. Additional diagnostic tools include CT scans, MR imaging studies, thyroglobulin (TG), and CEA measurements. In this specific case study patient's physical examination findings related to the thyroid gland were normal along with unremarkable results from a thyroid scan test evaluating both function and TG levels. There is ongoing debate regarding the necessity of surgery for this patient, the extent of the operation, and whether cervical lymph node dissection is warranted. Although OTPC is generally associated with low invasiveness and a benign biological behavior, clinical practice has revealed cases of lymph node metastasis, distant metastasis, and even mortality [22]. It is commonly accepted that unilateral thyroid gland and isthmus resection can be considered for solitary masses in OTPC; however, if there is tumor invasion into the capsule or multiple lesions are present, total thyroidectomy along with prophylactic cervical lymph node dissection may be performed. In this case, the patient underwent total thyroidectomy and prophylactic cervical lymph node dissection. Postoperative pathology confirmed papillary thyroid carcinoma as a multifocal lesion with capsular invasion; nevertheless, no evidence of cervical lymph node metastasis was observed. Subsequent postoperative radiiodine therapy and TSH
inhibition therapy were administered. Currently, there have been no significant signs of recurrence or metastasis in the patient.

CONCLUSION

Submandibular solid-cystic mass as the first and sole manifestation of occult thyroid papillary carcinoma is relatively rare. For the neck masses fine needle aspiration is necessary.


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