

# World Journal of *Clinical Cases*

*World J Clin Cases* 2023 October 16; 11(29): 6974-7260



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**RESPONSIBLE EDITORS FOR THIS ISSUE**

Production Editor: Hua-Ge Yin; Production Department Director: Xiang Li; Editorial Office Director: Jin-Lei Wang.

**NAME OF JOURNAL**

*World Journal of Clinical Cases*

**ISSN**

ISSN 2307-8960 (online)

**LAUNCH DATE**

April 16, 2013

**FREQUENCY**

Thrice Monthly

**EDITORS-IN-CHIEF**

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

**EDITORIAL BOARD MEMBERS**

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

**PUBLICATION DATE**

October 16, 2023

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<https://www.wjgnet.com/bpg/GerInfo/288>

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<https://www.wjgnet.com/bpg/gerinfo/208>

**ARTICLE PROCESSING CHARGE**

<https://www.wjgnet.com/bpg/gerinfo/242>

**STEPS FOR SUBMITTING MANUSCRIPTS**

<https://www.wjgnet.com/bpg/GerInfo/239>

**ONLINE SUBMISSION**

<https://www.f6publishing.com>



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

Geng-Yu Chen, Tong Li

**Specialty type:** Medicine, research and experimental

**Provenance and peer review:** Unsolicited article; Externally peer reviewed.

**Peer-review model:** Single blind

**Peer-review report's scientific quality classification**

Grade A (Excellent): 0  
Grade B (Very good): 0  
Grade C (Good): 0  
Grade D (Fair): 0  
Grade E (Poor): 0

**P-Reviewer:** Nambi G, Saudi Arabia

**Received:** August 28, 2023

**Peer-review started:** August 28, 2023

**First decision:** September 4, 2023

**Revised:** September 11, 2023

**Accepted:** September 26, 2023

**Article in press:** September 26, 2023

**Published online:** October 16, 2023



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### Abstract

#### BACKGROUND

Occult thyroid papillary carcinoma (OTPC) is typically characterized by initial presentation with cervical lymph node metastasis and can be detected through ultrasound. However, the initial and sole manifestation was a submandibular solid-cystic mass. High-frequency ultrasound, enhanced multislice computed tomography (CT) scan, and thyroid function tests revealed no abnormalities, which is relatively uncommon.

#### CASE SUMMARY

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

#### CONCLUSION

The presentation of a submandibular solid-cystic mass as the primary and solitary indication of OTPC is relatively uncommon. Fine needle aspiration is advised for evaluating neck masses.

**Key Words:** Occult thyroid carcinoma; Submandibular mass; Ultrasound; Fine needle aspiration; Case report

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**Core Tip:** Occult thyroid papillary carcinoma is rare. However, submandibular solid-cystic mass was the initial and sole manifestation, ultrasound and computed tomography scan 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.

**Citation:** Chen GY, Li T. Submandibular solid-cystic mass as the first and sole manifestation of occult thyroid papillary carcinoma: A case report. *World J Clin Cases* 2023; 11(29): 7253-7257

**URL:** <https://www.wjgnet.com/2307-8960/full/v11/i29/7253.htm>

**DOI:** <https://dx.doi.org/10.12998/wjcc.v11.i29.7253>

## 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% to 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 [7]. Occult thyroid papillary carcinoma (OTPC) refers to any PTC 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 computed tomography (CT) scanning, or 131I whole-body follow-up scan (131I WBS) [8-10]. However, some cases of OTPC remain undetected [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 she had noticed 2 mo earlier.

### History of present illness

A 24-year-old Chinese female, who studied at a university in Shandong Province, presented to the clinic in June 2019 with a right submandibular mass that she had noticed 2 mo earlier. Clinical examination revealed a 2-cm, nontender, movable solid-cystic mass in the submandibular region, with no palpable thyroid mass observed. Ultrasonography revealed a 2.0 cm × 1.1 cm solid-cystic mass in the right submandibular region, and the thyroid gland showed 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 PTC. The pathology revealed metastatic carcinoma in lymphoid tissue with morphology consistent with thyroid papillary carcinoma metastasis (Figure 1).

### History of past illness

Without special past illness.

### Personal and family history

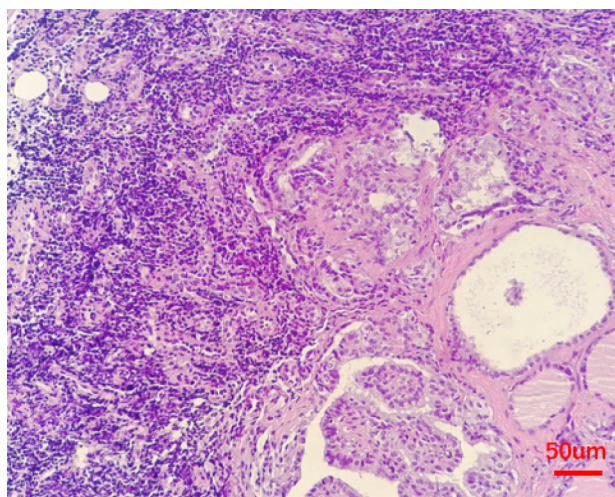
Without special personal and family history.

### Physical examination

Clinical examination revealed a 2-cm, nontender, movable solid-cystic mass in the submandibular region, with no palpable thyroid mass observed.

### Laboratory examinations

Laboratory examinations were all at normal levels.



DOI: 10.12998/wjcc.v11.i29.7253 Copyright ©The Author(s) 2023.

**Figure 1** Pathological reveal: Metastatic carcinoma in lymphoid tissue, combined with morphology in line with thyroid papillary carcinoma metastasis (hematoxylin and eosin stain, 200 ×).

### Imaging examinations

Ultrasonography demonstrated a 2.0 cm × 1.1 cm solid-cystic mass in the right submandibular region while revealing no abnormalities in the thyroid gland (Figure 2). US-guided fine-needle aspiration (FNA) exhibited papillary tumor cell clusters consistent with PTC.

## FINAL DIAGNOSIS

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 the right side showed no tumor metastasis but chronic inflammation.

## TREATMENT

The patient was treated by total thyroidectomy and right neck lymph node dissection. Postoperative radioiodine therapy and thyrotropin (TSH) inhibition therapy were administered after surgery.

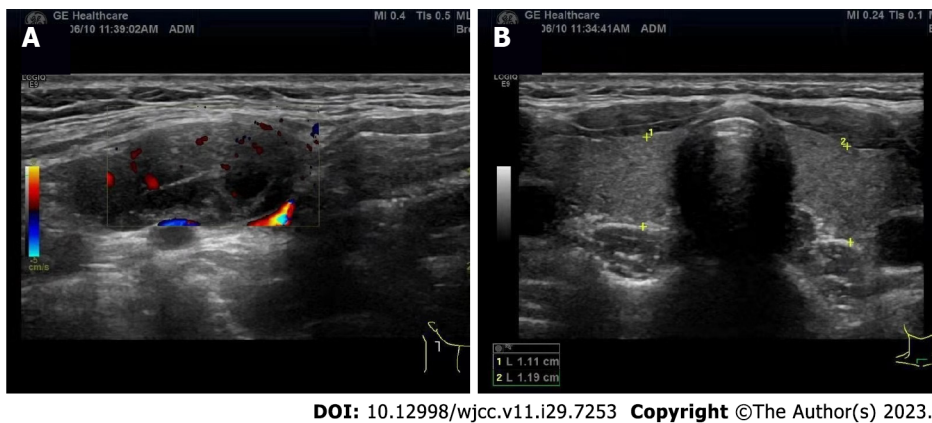
## OUTCOME AND FOLLOW-UP

The patient has been followed up for 2 years without significant recurrence.

## DISCUSSION

Regarding the progressive enlargement of masses in the submandibular region, especially in young patients, we strongly recommend heightened vigilance. Although most are benign lumps, such as submandibular lymphadenitis, submandibular cysts, and hypoglossal adenitis, reported cases of malignant or metastatic lymph nodes have been documented. Ethar reported a case where a right submandibular mass was diagnosed as mammary analogue secretory carcinoma through fine needle aspiration[12]. Chen *et al*[13] 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. Miles demonstrated that a right submandibular mass, confirmed on histological analysis following excision of the right submandibular gland, was multinodular adenomatous oncocyctic hyperplasia[14]. Performing high-frequency ultrasound to evaluate 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, 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





**Figure 2 Ultrasonography reveal.** A: 2.0 cm × 1.1 cm solid-cystic mass with clear border and regular shape, and the separation can be seen inside; B: The thyroid gland has no abnormalities.

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 FNA was performed which showed papillary tumor cell clusters consistent with PTC. Further surgical intervention indicated lymph node metastasis from PTC. Some studies have shown that PTCs frequently undergo cystic transformation, 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[11,18]. High-frequency ultrasound serves as the gold standard for diagnosing thyroid cancer by enabling detection of lesions as small as 3 mm[19]. Thyroid cancer exhibits irregular margins, hypoechogenicity, and microcalcifications. Malignant nodules demonstrate significantly greater hardness than benign ones. Real-time shear wave elastography plays a crucial role in differentiating between benign and malignant nodules[20]. Additional diagnostic tools include CT scans, magnetic resonance imaging studies, thyroglobulin (TG), and carcinoembryonic antigen measurements. In this specific case study, the 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[21]. 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 PTC as a multifocal lesion with capsular invasion. Nevertheless, no evidence of cervical lymph node metastasis was observed. Subsequent postoperative radioiodine therapy and TSH inhibition therapy were administered. Currently, there have been no significant signs of recurrence or metastasis in the patient.

## CONCLUSION

The occurrence of a submandibular solid-cystic mass as the initial and sole manifestation of OTPC is relatively uncommon. Fine needle aspiration is necessary for evaluating neck masses.

## FOOTNOTES

**Author contributions:** Chen GY collected the clinical data; Li T wrote the manuscript and analyzed the data.

**Informed consent statement:** Informed written consent was obtained from the patient for publication of this report and any accompanying images.

**Conflict-of-interest statement:** The authors declare that they have no conflict of interest.

**CARE Checklist (2016) statement:** The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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**S-Editor:** Qu XL

**L-Editor:** A

**P-Editor:** Qu XL

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