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Synchronous Occurrence of Thyroglossal Duct Cyst-Derived Papillary Carcinoma and Papillary Thyroid Carcinoma: A Case Report and Literature Review

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Abstract

The thyroglossal duct (TGD) normally regresses; when it persists, a thyroglossal duct cyst (TGDC) may form. Malignancy in a TGDC is rare. Papillary thyroid carcinoma (PTC) is the most common differentiated thyroid cancer, and the concurrent occurrence of papillary carcinoma of the TGDC (TGDC-PC) and PTC is uncommon. We report one such synchronous case. A young woman presented with neck discomfort and a foreign-body sensation. Imaging revealed a TGDC; the thyroid appeared normal. Intraoperative frozen-section examination confirmed TGDC-PC. On this basis, she underwent a Sistrunk procedure, total thyroidectomy (TT), and regional lymph node dissection. Postoperative pathology identified PTC with lymph node metastasis. When papillary carcinoma arises in a TGDC, a hidden thyroid lesion should be suspected despite normal imaging, and treatment should proceed accordingly.

Categories: Otolaryngology, General Surgery, Oncology
Keywords: mechanism, multidisciplinary collaboration, papillary thyroid carcinoma, pathological diagnosis, surgery, thyroglossal duct cyst, thyroid gland

Introduction

Thyroglossal duct cysts (TGDCs) occur in 7% of the population, with malignant transformation occurring in 0.7% to 1.0% of cases [1,2]. Papillary thyroid carcinoma (PTC) accounts for approximately 85% of differentiated thyroid malignancies [3]. The concurrent occurrence of TGDC and PTC is exceedingly uncommon. The pathogenesis of papillary carcinoma of the TGDC (TGDC-PC) remains poorly defined, and its relationship to PTC has received little attention. There is no consensus on the diagnosis and management of synchronous thyroglossal duct cyst carcinoma (TDCC) and thyroid carcinoma. Distinguishing a primary tumour from a metastasis is difficult, and subsequent treatment remains controversial. For isolated TDCC, the Sistrunk procedure is the standard treatment; however, in this patient population, performing only the Sistrunk procedure may carry a risk of missing thyroid lesions.

Case Presentation

History and physical examination

Chief Complaint

A 37-year-old female patient presented with a foreign body sensation and discomfort in the neck for two years, with symptoms worsening over the past two days.

Past Medical History

The patient had no prior thyroid disorders and denied any history of radiation exposure.

Physical Examination

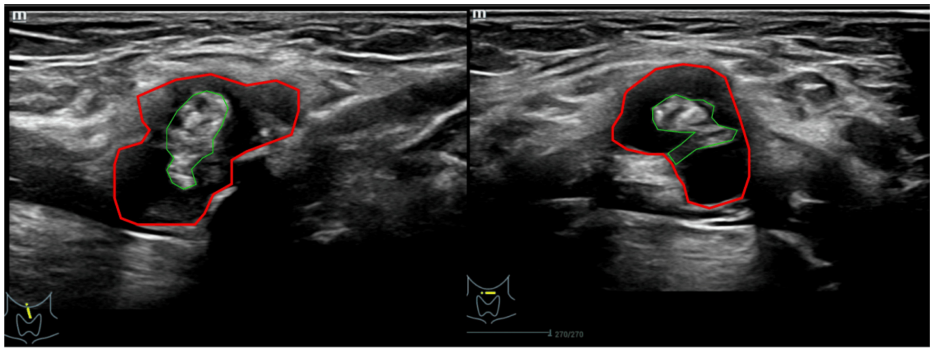
A 2 × 2 × 1.5 cm non-tender mass was palpable in the anterior midline of the neck, at the level of the hyoid bone. The lesion had a moderately firm consistency, a smooth surface, and well-defined borders, and retraction was observed when the patient protruded her tongue. No thyroid gland enlargement, intralobar nodules in either thyroid lobe, or obviously enlarged cervical lymph nodes were identified during physical examination.

Auxiliary examinations

Imaging

Neck ultrasound revealed no thyroid abnormalities (Figure *1*), but a mixed cystic-solid mass was detected subcutaneously along the midline of the neck (Figures *2A*-*2B*). Contrast-enhanced neck MRI demonstrated an abnormal signal in the thyrohyoid region, suggestive of a neoplastic lesion. The lesion exhibited iso- to slightly hyperintense signal on T1-weighted imaging (T1WI), heterogeneous slight hyperintensity on T2-weighted imaging (T2WI), and heterogeneous enhancement on post-contrast scans. Multiple cervical lymph nodes were visualised (Figures *3A*-*3B*).

FIGURE 1: Neck ultrasound revealed a symmetrical thyroid gland with a regular shape and heterogeneous parenchymal echogenicity. No obvious masses were observed.



A B

FIGURE 2: Neck ultrasound revealed a mixed cystic-solid mass located subcutaneously along the midline of the neck. Red markers indicate the cystic components, while green markers indicate the solid components within the cyst.

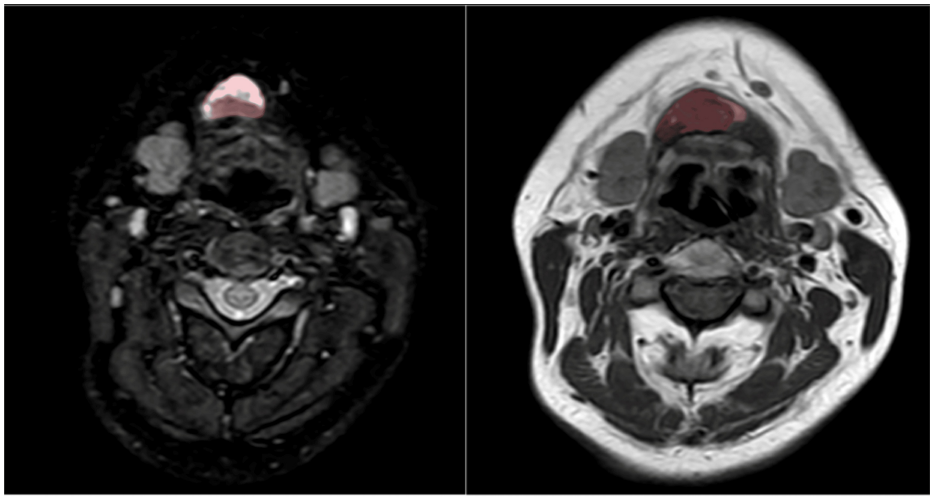


FIGURE 3: Contrast-enhanced MRI of the neck revealed an abnormal signal in the thyrohyoid region, appearing isointense to mildly hyperintense on T1-weighted imaging (T1WI) (A) and heterogeneously mildly hyperintense on T2-weighted imaging (T2WI) (B).

Laboratory Tests

Thyroid function tests (thyroid-stimulating hormone (TSH), free triiodothyronine (FT3), and free thyroxine (FT4)), thyroglobulin (Tg), CBC, blood biochemistry, and serum electrolyte levels were all within normal limits. The anti-thyroid peroxidase antibody level was 13.07 IU/mL, and the 25-hydroxyvitamin D level was 17.18 ng/mL.

Surgical procedure

The patient declined fine-needle aspiration biopsy (FNAB) before surgery. Intraoperatively, a 2 × 2 × 1.5 cm cystic-solid mass was identified beneath the hyoid bone, with enlarged level VI lymph nodes; the thyroid gland showed no distinct mass. We then performed the Sistrunk procedure, excising the cyst, the middle portion of the hyoid bone, and part of the tongue base. Frozen-section examination showed papillary thyroid carcinoma components in the cyst wall of the TGDC. After intraoperative consultation with the family, we biopsied a level VI lymph node, which confirmed metastasis. To exclude occult thyroid cancer, we proceeded with total thyroidectomy (TT) and therapeutic level VI lymph node dissection.

Pathological examination

Intraoperative frozen-section examination confirmed PTC in the TGDC wall. Postoperative pathological examination of the neck mass specimen revealed a cystic mass. Papillary structures with calcifications were observed on the cyst wall, and the nuclei were enlarged, with a ground-glass appearance and visible nuclear grooves. Papillary thyroid carcinoma was identified on the inner cyst wall, with the carcinoma focus measuring approximately 5 mm in its maximum dimension (Figure *4*). A papillary thyroid carcinoma measuring 0.2 × 0.2 × 0.2 cm was found in the right thyroid lobe without invasion of the thyroid capsule (Figure *5*). The left thyroid lobe showed nodular goitre, and level VI cervical lymph node metastasis was detected in 1 of 21 lymph nodes (1/21), with the maximum diameter of the metastatic lesion measuring 0.4 mm (Figure *6*).

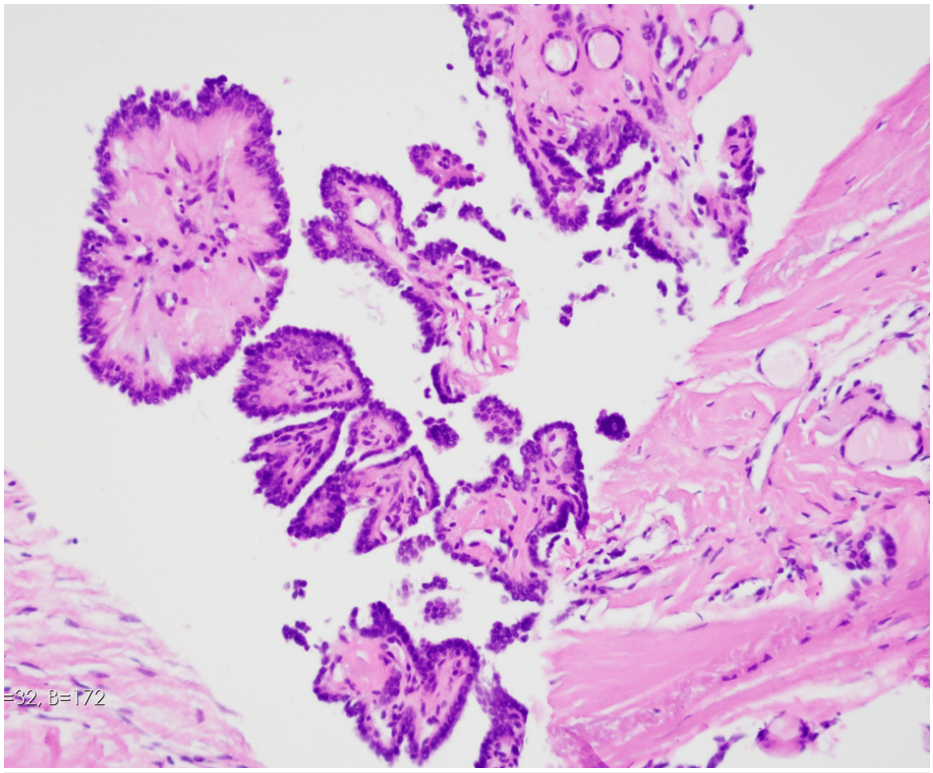


FIGURE 4: The tumour grows in a papillary pattern within the TGDC capsule wall. Its cells show large, densely packed nuclei with ground-glass chromatin, visible nuclear grooves, and intranuclear pseudoinclusions (H&E staining, magnification ×100).

TGDC:Thyroglossal duct cyst.

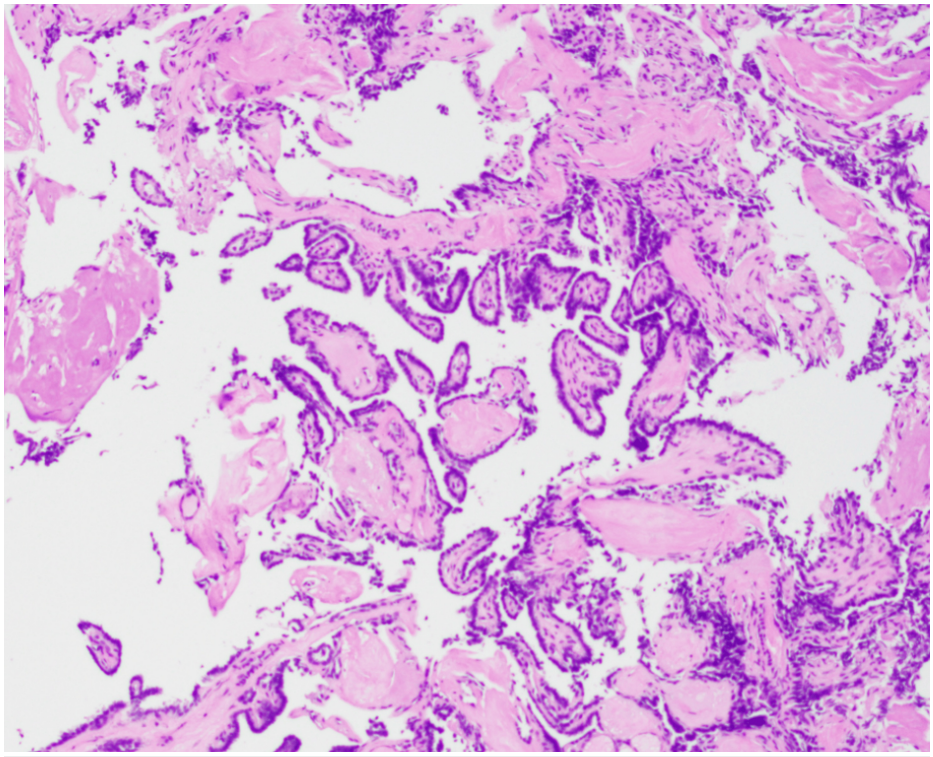


FIGURE 5: Papillary carcinoma is visible within the thyroid gland, exhibiting an infiltrative growth pattern (H&E staining, magnification ×40).

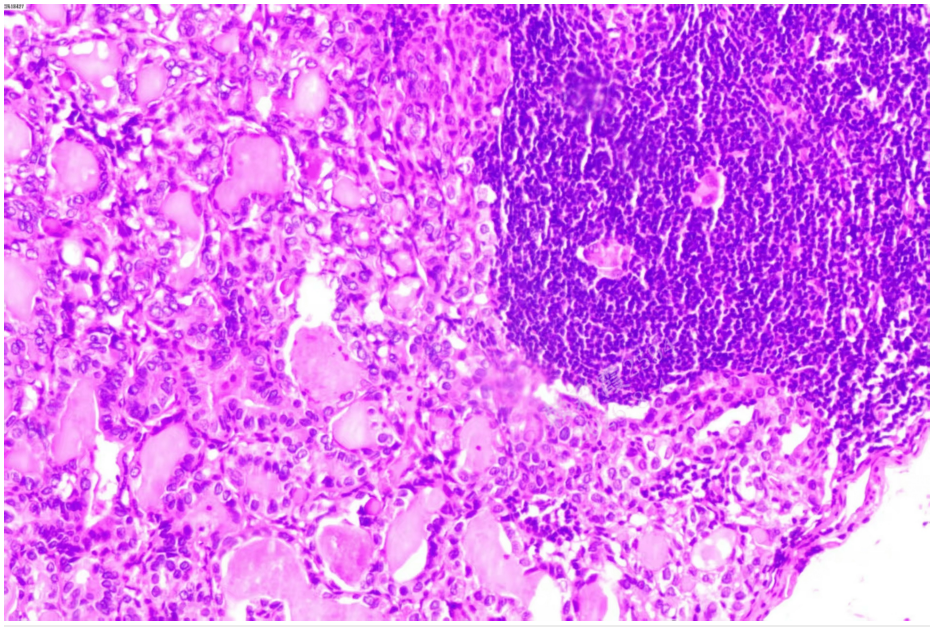


FIGURE 6: The lymph node contained focal papillary thyroid carcinoma, confirming nodal metastasis (H&E staining, magnification ×100).

Discussion

Foregut endoderm invaginates at the foramen cecum to form the thyroid gland, which then migrates from the tongue base to the anterior neck. The TGD normally regresses but persists in roughly 7% of the adult population, typically presenting as a TGDC. TGDCs occur more often in female children. Over half of patients undergoing surgery for TGDC had functionally normal ectopic thyroid tissue [1,4]. Malignant

transformation in TGDC is uncommon, occurring in 0.7% to 1.0% of cases. Since Brentano's description in 1911, only about 300 cases have been reported in the English-language literature. Papillary carcinoma accounts for 75-80% of primary TDCCs. The remaining subtypes include mixed papillary-follicular carcinoma (7%), squamous cell carcinoma (5%), follicular carcinoma (1.7%), and Hürthle cell or undifferentiated carcinoma (0.9%) [2].

Pathophysiology controversy

Limited data exist on the pathophysiological features of papillary carcinoma arising in TGDC. Two mechanisms may explain PTC development in TGDC: malignant transformation of residual thyroid tissue within the duct or metastatic seeding from an occult thyroid primary [5]. TGD-associated malignancy requires malignant lesions in TGD remnants, with intact normal thyroid parenchyma. In this patient, synchronous PTC involved both the TGDC and the thyroid, a finding outside the standard definition of TGD-related carcinoma. A prior study of 242 patients with a history of TGD identified TGDC in 217 individuals; ectopic thyroid tissue was found in the cyst wall in 68 cases (31.3%). Of the 18 papillary carcinoma cases in that cohort, 7 met the criteria for TGD-associated papillary carcinoma; the remaining 11 could not be distinguished from primary pyramidal lobe carcinoma, Delphian node metastasis, or TGD-originated carcinoma [6]. Using multiple thyroid markers, we immunophenotyped TGDC samples. Staining was positive for TTF-1 and other thyroid antigens but negative for calcitonin. These findings confirm the presence of thyroid follicular cells in the TGD cyst wall [7]. A retrospective study reported discordant histopathological tumour types between thyroid lesions and TDCC in 6.4% of cases [8]. Synchronous thyroid carcinoma and TDCC appear to be genotypically independent, supporting a primary origin for each [9].

In one reported case, imaging and fine-needle aspiration (FNA) cytology suggested TGDC but also revealed features of papillary carcinoma. Postoperative histopathology of the resected lesion confirmed cystic degenerative lymph node metastasis of papillary carcinoma. Thyroidectomy was performed based on the pathological findings, with a final diagnosis of lymphocytic thyroiditis and no papillary carcinoma lesions [10]. The coincidence rate of orthotopic thyroid carcinoma with TDCC is 34.6% across reported series. Among synchronous cases, up to 90.0% show PTC in both TDCC and thyroid tissues; medullary carcinoma coexists in rare instances [8]. Ectopic thyroid parenchyma appears in two-thirds of TGDCs, while parafollicular cells are absent from these heterotopic tissues [11]. Since medullary thyroid carcinoma (MTC) arises from parafollicular cells, this histological difference lends support to the possibility that TDCC originates from occult thyroid metastasis, although confirmatory evidence is lacking. It is impossible to distinguish the sequence of onset of thyroid carcinoma and TDCC in the present case.

Diagnostic challenges

Neck masses are commonly evaluated using ultrasound, CT, and MRI. Solid components within a TGDC on imaging should raise suspicion for TDCC. Typical CT findings include mural nodules (dense or progressively enlarging), intracystic calcification, or both [12]. FNAB is safe, well tolerated, and cost-effective for head and neck lesions; repeat sampling can provide sufficient material [4]. FNA has excellent diagnostic accuracy for various tissue lesions, reported at 99.2% for head and neck masses. For cystic neck lesions, preoperative FNA correlates closely with postoperative pathology [13]. FNA samples from TGDCs typically yield cystic fluid containing macrophages, neutrophils, and sparse epithelial cells in a mucoid or proteinaceous background. Squamous cells predominate among the epithelial components; ciliated columnar cells are occasionally seen. Ectopic thyroid tissue within the fibrous cyst wall is difficult to sample by routine FNA [7]. Preoperative FNA diagnosis of TGDC malignancy is uncommon; the reported true-positive rate is 53%, with a false-negative rate of 47% [12,14]. Although only about 60% sensitive for TDCC, FNA remains useful for cystic lesions with solid areas, especially when thyroid nodules coexist. Multiple studies advise three or more passes, with pathology support, to improve diagnostic yield [14]. Preoperative discussion among surgeons, radiologists, and pathologists is important. Thorough imaging review guides surgical decisions, and careful preoperative assessment directs the procedure.

Treatment strategy

Core Procedure

The Sistrunk procedure combined with total thyroidectomy (TT) may be considered for patients with concomitant thyroid cancer. Controversy persists over whether TDCC warrants total thyroidectomy or neck dissection in addition to the Sistrunk procedure. The Sistrunk procedure, involving removal of the cyst and central portion of the hyoid bone, is the minimum procedure needed to reduce recurrence [15]. Whether to add TT or perform cervical lymph node dissection remains debated. Some recommend TT with the Sistrunk procedure even with a normal preoperative workup, citing the risk of secondary thyroid malignancy; prophylactic lymph node dissection is an option in selected cases [16]. One classification system assigns low-risk status to patients aged <45 years with a tumour diameter of <4.0 cm, no nodal or distant metastases, and no prior neck irradiation. High-risk status is assigned to patients aged ⩾45 years with a tumour diameter of ⩾4.0 cm, nodal or distant metastases, prior irradiation, or concurrent thyroid nodules. According to this framework, high-risk patients undergo the Sistrunk procedure plus TT, postoperative radioactive iodine (RAI) therapy when appropriate, and regional lymph node dissection if indicated [17,18]. Our patient, who met the high-risk criteria, underwent the Sistrunk procedure with TT and regional lymph

node dissection; resection was complete. RAI was declined postoperatively; TSH suppression alone was used. First-year surveillance at three-month intervals included thyroid USG, thyroid function tests (TSH, FT3, and FT4), and serum Tg. All values remained within the target ranges; no imaging or biochemical findings suggested recurrence or metastasis.

Prognosis and follow-up

For TDCC, especially the PTC subtype, the prognosis is good, with a 10-year survival rate of 95-100%, surpassing that of primary PTC. The simultaneous presentation of thyroid carcinoma, TDCC, and nodal metastasis is uncommon. Over a mean follow-up of 41.5 months, the reported mortality rate was 0.95%, and the recurrence rate was 7.4%, compared with the previously reported rate of 4.3% [8]. Distant metastases occurred in 1.4% of cases, and locoregional spread occurred in 20% [8]. Lateral neck involvement was observed in 15% of cases and central compartment involvement in 5.6%, consistent with earlier findings that lymphatic spread often follows the superior thyroid pedicle to the jugular chain [19]. The nodal metastasis rate for primary PTC, in contrast, is 89.8% [20]. Whether this patient's thyroid carcinoma metastasised to the TGD via lymphatics remains unconfirmed.

In this case, negative thyroid imaging did not exclude occult thyroid lesions or lymph node metastasis. Intraoperative frozen-section examination also proved useful in deciding whether to extend surgery to total thyroidectomy and therapeutic lymph node dissection. This sequence may serve as a practical reference for similar presentations and support more individualised treatment planning.

Conclusions

TDCC is a rare malignancy with a generally favourable prognosis. Preoperative characterisation of the mass and identification of concomitant lesions help inform surgical planning. Evidence on concurrent thyroid cancer and TDCC remains limited. In TDCC, negative preoperative thyroid imaging does not exclude occult lesions; intraoperative frozen-section examination should therefore be integrated into surgical decision-making. For patients meeting high-risk criteria, the Sistrunk procedure combined with TT and regional lymph node dissection may be considered, although the extent of surgery should be individualised. Long-term follow-up with thyroid USG and thyroid function tests is advised to detect recurrence or metastasis early.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Jie Chen, Cai Xia Lai, Qiao Lian Deng, Hong Ying Che

Acquisition, analysis, or interpretation of data: Jie Chen, Cai Xia Lai, Qiao Lian Deng

Drafting of the manuscript: Jie Chen, Cai Xia Lai, Qiao Lian Deng

Critical review of the manuscript for important intellectual content: Jie Chen, Hong Ying Che

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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