Case Report

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Papillary Carcinoma of Thyroglossal Duct Cyst with Ectopic Thyroid: A Case Report

Radiology Section

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ABSTRACT

Papillary carcinoma arising from a Thyroglossal Duct Cyst (TGDC) is an uncommon malignancy, accounting for less than 1% of all TGDCs. Imaging plays a crucial role in early detection, characterisation, and guiding surgical management. We report a case of papillary carcinoma developing in a TGDC in a 27-year-old male who presented with a gradually enlarging anterior midline neck swelling, recently associated with pain. There were no symptoms of dysphagia, hoarseness, or thyroid dysfunction. Ultrasonography revealed a well-defined predominantly cystic lesion with internal septations and echogenic components showing vascularity on colour Doppler. Contrast-Enhanced Computed Tomography (CECT) of the neck demonstrated a thick-walled cystic lesion below the hyoid bone with enhancing internal septa and solid nodular areas, suggesting malignant transformation. Incidentally, a separate enhancing nodule was observed at the base of the tongue, consistent with ectopic (lingual) thyroid tissue, along with a small orthotopic thyroid gland. No cervical lymphadenopathy was noted. The patient underwent Sistrunk's procedure, and histopathological evaluation confirmed papillary carcinoma arising within the TGDC. The postoperative period was uneventful. This case highlights the significance of meticulous preoperative imaging in identifying both malignant features within a TGDC and the presence of additional thyroid tissue along the thyroglossal tract.

Keywords: Contrast-enhanced CT, Lingual thyroid, Sistrunk procedure

CASE REPORT

A 27-year-old male presented with swelling in the right anterior midline of the neck for two years, which recently started increasing in size and was associated with pain. He had no history of dysphagia, hoarseness, hypothyroidism, hyperthyroidism, fever or radiation exposure. Other systemic findings were unremarkable.

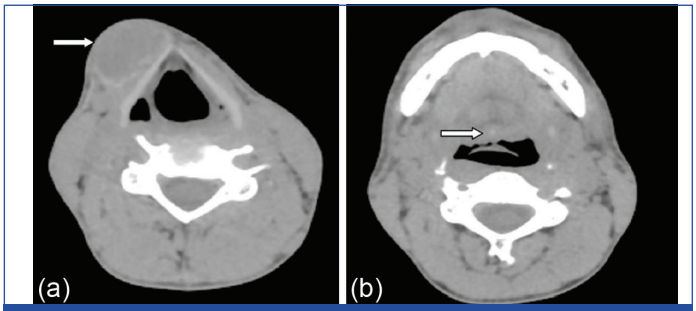
On examination, a well-defined, oval swelling measuring approximately 4×3 cm was noted in the right anterior midline of the neck. The swelling was firm in consistency, tender on palpation, and moved upwards with deglutition and tongue protrusion. The overlying skin appeared normal, and no cervical lymphadenopathy was detected.

Routine blood investigations, including the complete blood count, renal and liver function tests, were within normal limits. Thyroid function tests demonstrated values within the reference range- T3: 1.6 ng/mL (normal: 0.8-2.0 ng/mL), T4: 9.2 µg/dL (normal: 5-12 µg/dL), and Thyroid Stimulating Hormone (TSH): 2.8 µIU/mL (normal: 0.4-4.0 µIU/mL)- indicating a euthyroid state.

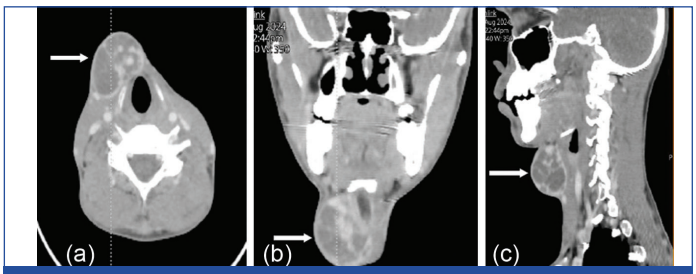
Ultrasonography of the neck showed a well-defined, predominantly cystic lesion measuring approximately 42(TR)×25 (AP)×40 (CC) mm to the right of the midline, with internal septae and an echogenic component showing vascularity on colour Doppler.

Contrast-enhanced Computed Tomography (CECT) of the neck was performed, which showed a well-defined, thick-walled, hypodense, predominantly cystic lesion measuring approximately 44(TR)×28(AP)×43(CC)mm in the subcutaneous plane at the anterior midline towards the right side, below the level of the hyoid bone. On post-contrast study, there was a thick-walled enhancing lesion with multiple internal septa and nodular solid areas. The remaining visualised structures of the neck were unremarkable. CECT also showed a well-defined nodular enhancing area measuring approximately 6×7 mm, noted in the posterior aspect of the tongue, suggestive of lingual thyroid [Table/Fig-1-3]. Both ultrasonography and computed tomography revealed no abnormal cervical lymph nodes.

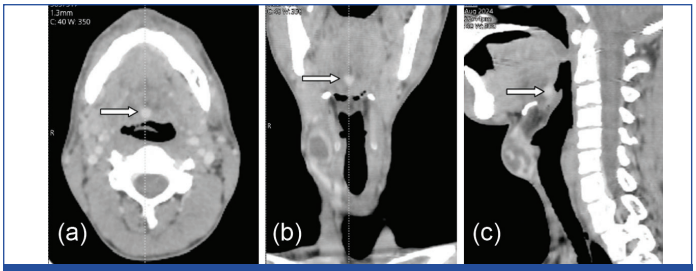
Based on ultrasonography and computed tomography imaging findings, a provisional diagnosis of papillary carcinoma of the thyroglossal duct was made. Patient underwent Sistrunk surgery; the lesion and the tract were removed in total, along with a part of the body of the hyoid bone. The patient's postoperative period was uneventful. The final histopathological results revealed papillary fronds with fibrovascular cores lined by cells having nuclear clearing and grooves, confirming papillary carcinoma arising in a TGDC. At one-year follow-up, the patient remained disease-free with normal thyroglobulin levels and no recurrence on neck ultrasound.



[Table/Fig-1]: Plain CT of the neck, (a) Axial CT at C7 level showing a well-defined hypodense lesion with a hyperdense capsule right of midline, in subcutaneous neck planes limited posterior-laterally by thyroid cartilage. (b) Axial CT at the epiglottis level showing a well-defined isodense lesion at the base of the tongue anterior to the pre-epiglottic space — suggestive of ectopic thyroid tissue.



[Table/Fig-2]: (a–c) Axial, coronal, and sagittal CECT neck images showing an enhancing thick-walled cystic lesion with internal septae and nodular solid areas right of midline below the hyoid bone, extending from C5 to C8.



[Table/Fig-3]: (a–c) Axial, coronal, and sagittal CECT neck images showing a well-circumscribed, avidly enhancing mass at the base of the tongue anterior to the pre-epiglottic space — suggestive of ectopic (lingual) thyroid.

DISCUSSION

The thyroid gland originates at the foramen caecum and descends to its normal pretracheal position, leaving behind the thyroglossal duct during embryogenesis. Failure of involution of this duct can result in ectopic thyroid tissue or a TGDC; TGDCs are reported in up to 7% of adults [1,2].

TGDCs are usually benign; however, malignant transformation occurs in approximately 1% of cases [3]. Papillary thyroid carcinoma is the most common histological subtype identified in TGDC malignancies [4,5]. The origin of carcinoma in TGDCs remains controversial, with theories including *de novo* malignant transformation of ectopic thyroid tissue or spread from an occult primary thyroid carcinoma [4].

Imaging plays a crucial role in raising suspicion for malignancy within a TGDC. Features such as mural nodules, thickened cyst walls, internal septations, and vascular solid components are suggestive of malignant change [5,6]. In the present case, ultrasonography and CECT demonstrated a thick-walled cystic lesion with enhancing internal septae and nodular components, consistent with these malignant imaging characteristics.

An unusual and clinically important finding in our patient was the co-existence of ectopic lingual thyroid tissue along with a small orthotopic thyroid gland. Ectopic thyroid tissue most commonly occurs at the base of the tongue and may represent the only functioning thyroid tissue in some individuals, making its preoperative identification essential [2,7].

Recent literature continues to emphasise the rarity of papillary carcinoma arising in TGDCs. A recent case reported by Fekadu D et al., described papillary carcinoma within a TGDC without associated ectopic thyroid tissue [8]. Plaza CPR et al., reported a series of TGDC carcinomas emphasising the importance of complete surgical excision via the Sistrunk procedure, sometimes supplemented by thyroidectomy in patients with coexisting thyroid abnormalities [1]. Gomez-Álvarez LR et al., described multiple cases of TGDC carcinoma with variable imaging findings and stressed the unpredictable extent of the disease [6]. Maleki N et al., presented a case similar to ours, in which papillary carcinoma arose within a TGDC [5]; however, the co-existence of a lingual thyroid and orthotopic thyroid gland, as in our patient, has rarely been documented. Sistrunk's procedure remains the treatment of choice for TGDC carcinoma, with additional thyroidectomy reserved for selected cases based on thyroid involvement or high-risk features. Prognosis is generally excellent following complete surgical excision [3,7].

Imaging Differentials

Based on the clinical history and examination findings, we conclude that the lesion is congenital and communicates with the base of the tongue. Given the location of neck cystic lesions, the differential diagnoses include midline and lateral lesions. Midline lesions, located within 2 cm of the midline, include a ranula, a thyroglossal cyst, and a dermoid cyst. An upper ranula is ruled out, as these typically occur at the base of the tongue. A midline thyroglossal cyst is characterised by upward movement with the hyoid bone upon tongue protrusion. A lower dermoid cyst is less likely, as ultrasonography typically shows dermoid cysts as homogeneously hyperechoic, whereas a thyroglossal cyst is usually hypoechoic and may contain internal echoes; differentiation can be challenging if a dermoid cyst is located near the hyoid bone. Lateral neck lesions include branchial cleft cysts, which are ruled out as they usually occur along the anterior border of the sternocleidomastoid, as well as lymphangiomas and vascular malformations. Solid neck lesions that should also be considered include lymph nodes, thyroid lesions, and ectopic thymus.

CONCLUSION(S)

This case reinforces the need for thorough preoperative imaging to confirm the presence of orthotopic thyroid tissue and identify ectopic thyroid locations. Such evaluation helps prevent postoperative hypothyroidism if the ectopic tissue is the only functioning thyroid source. A combination of ultrasonography and CT remains the cornerstone for diagnosis and surgical planning.

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