

Papillary Thyroid Carcinoma Arising in Thyroglossal Cyst: A Report of Two Cases

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Abstract

Papillary carcinoma arising in a thyroglossal duct cyst is a rare entity, accounting for approximately 1% of all thyroglossal cyst cases. Clinically, it often mimics a benign cystic lesion, making preoperative diagnosis difficult. In most instances, the malignancy is discovered incidentally during histopathological evaluation following surgical excision. We report two cases of papillary carcinoma arising in thyroglossal duct cysts in female patients aged 38 and 37 years, respectively. In both cases, the carcinoma was identified only after routine surgical excision performed for clinically benign-appearing cysts. Histopathological examination revealed features characteristic of papillary thyroid carcinoma originating within the thyroglossal duct cyst. These cases highlight the importance of thorough preoperative evaluation, including high-resolution ultrasound, to assess for solid components, calcifications, or other suspicious features. Furthermore, extensive histological sampling of excised thyroglossal duct cysts—particularly those with solid areas—is essential to exclude the presence of malignancy.

Keywords: Papillary carcinoma; Histopathology; Thyroglossal Cyst; Malignancy; Adenocarcinoma.

Introduction

A thyroglossal duct cyst (TGDC) is the most common congenital anomaly associated with thyroid development. During embryogenesis, the thyroid gland descends from the foramen cecum at the base of the tongue to its definitive location anterior to the thyroid cartilage. This migration occurs along the thyroglossal duct, a transient embryonic structure that typically involutes between the 5th and 10th weeks of gestation. Failure of complete ductal involution can result in the persistence of epithelial remnants, which may give rise to a thyroglossal duct cyst. These remnants may manifest as a cyst, persistent duct, fistula, or ectopic thyroid tissue, all of which can predispose to the formation of a TGDC. [1]

The exact stimulus for thyroglossal duct cyst expansion remains unclear; however, it is believed that lymphoid tissue associated with the duct may undergo hypertrophy during regional infections, leading to occlusion of the tract and subsequent cyst formation. Many thyroglossal duct cysts remain clinically silent and go undetected. The earliest documented cases of tumors arising from thyroglossal duct remnants were reported by Brentano in 1911 and Uchermann in 1915. When symptomatic, patients typically present around the median age of 40, often with painless, asymptomatic midline neck masses. [2]

Thyroglossal duct cyst papillary carcinoma is a rare finding, typically diagnosed after surgery through histopathology. The possibility of malignancy originating from thyroglossal duct remnants is 1-2%. [3] Till date, around 260 cases have been described in the literature. Papillary carcinoma is the most common type, making up 85-92% of these malignancies. [4] It is followed by squamous cell carcinoma, which accounts for about 7% of cases, which tend to be more aggressive with a worse prognosis. In most of the cases, the diagnosis of the thyroglossal duct carcinoma is made after the histopathologic examination of the surgical specimen. [5]

Here we present a case series of two cases of papillary carcinoma of thyroglossal duct cyst in 38-year & 37-year-old females. Papillary carcinoma was detected after a routine surgical operation was done for a seemingly innocuous thyroglossal cyst.

Case Report

Case 1: A 38-year-old female came to Surgery OPD with a complaint of midline neck swelling. A diagnosis of thyroglossal cyst was made by physical examination and findings of USG. FNAC showed features of colloid goiter, which was later proven to be a false negative. After surgical removal of the mass, it was sent for histopathological examination. A specimen of thyroglossal duct submitted showed a cystic structure filled with colloid measuring 1.5x0.5 cm along with a grey-white solid area measuring < 1 cm. On microscopic examination, it showed an encapsulated tumor composed of cells arranged in papillae, follicles, and tubules. Pseudostratification of nuclei is seen at places. Cells are cuboidal to columnar in appearance and areas of focal calcification. Tumor cells are positive for TTF1 and CK19.

Case 2: A 37-year-old female came to Surgery OPD with a complaint of swelling in front of the neck for the past 5 months, which moves with deglutition and protrusion of the tongue. CECT neck showed the right lobe of the thyroid gland was enlarged with a few sub-centimetric size hypodense nodules. A well-defined cystic lesion of size 38 x 20 mm is seen along the anterior aspect of strap muscles suggestive of a thyroglossal cyst. Multiple sub-centimetric sized non-specific level 1a and 1d lymph nodes were seen. FNAC showed features consistent with a thyroglossal cyst. After the diagnosis was made, the patient was taken for thyroglossal cyst excision under general anesthesia. The excised sample was sent for histopathological examination. On histopathological examination, grossly it was a soft tissue piece measuring 4 x 2 cm. On cut section, a greyish-brown tumor is identified in the cyst. The cyst identified measured 1.5 x 1.5 cm, yellowish gelatinous material was oozing out, and a small friable growth of 0.5x0.5 cm. Microscopic sections from the cyst showed cells arranged in a papillary architecture with nuclear crowding, overlapping, and focal calcification. These cells showed immunopositivity for CK19 and TTF. A section from the rest of the cyst wall shows a lining of ciliated columnar epithelium with focally eroded epithelium with underlying chronic inflammation, and focal colloid-filled thyroid follicles are also observed in the cyst parenchyma.



Figure 1: Gross specimen of thyroglossal duct.

Discussion

Though thyroglossal duct cysts are a frequent congenital thyroid abnormality, cancer within these cysts is exceedingly rare. In the vast majority of cases (approximately 90%), these carcinomas arise from residual thyroid tissue within the cyst. [6] Papillary thyroid carcinoma is the dominant histological type, accounting for roughly 94% of cases, while squamous cell

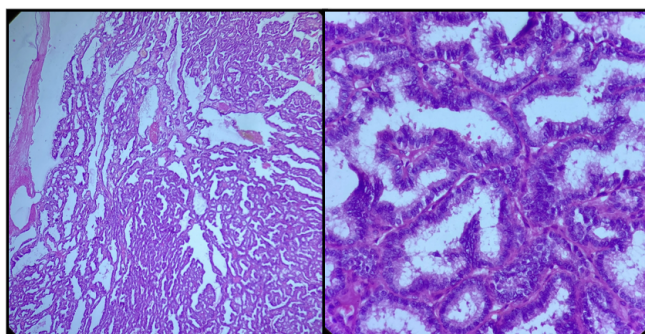


Figure 2: Photomicrograph showing tumor cells arranged in true papillae inside the cyst and nuclear features of papillary carcinoma with overcrowding of the nuclei and nuclear grooving (red arrow). (H&E, 200X and 400X).

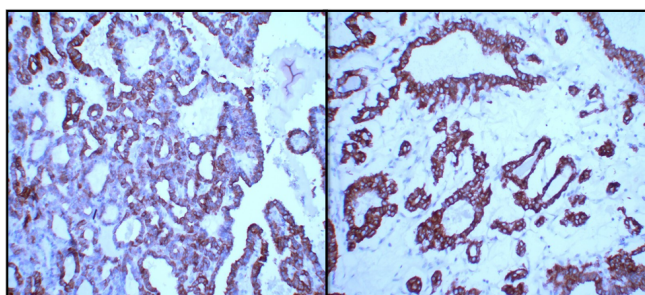


Figure 3: Photomicrograph showing IHC positivity for CK-19 (200X, 400X).

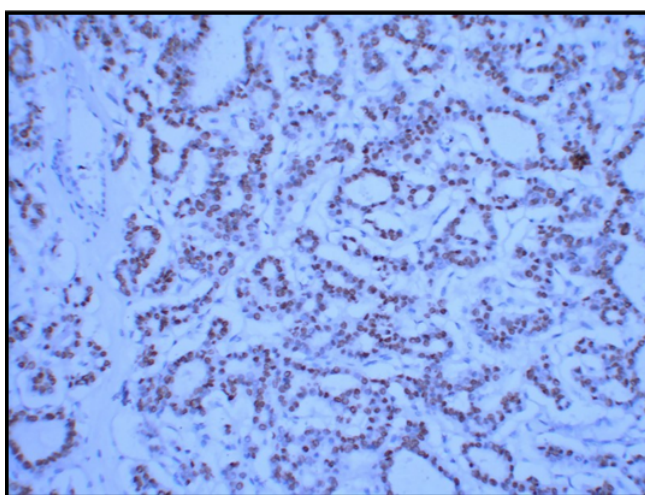


Figure 4: Photomicrograph showing tumor cells with positive immunoexpression for TTF (200X).

carcinoma occurs in less than 5%. [7] The precise cause of this malignancy remains elusive, and there are no reliable clinical or physical indicators that allow for preoperative diagnosis. [8]

The thyroid origin of thyroglossal duct adenocarcinomas is primarily explained by two main hypotheses, with the 'de novo' theory being the most widely accepted. This theory suggests that the carcinoma arises directly from ectopic thyroid tissue within the thyroglossal duct. Histopathological evidence supports this, with ectopic thyroid tissue identified in approximately 62% of cases. Further support for this theory comes from the absence of medullary carcinoma in these lesions, as medullary carcinoma originates from parafollicular (C) cells, which are not present in ectopic thyroid tissue derived from follicular cells. [9]

An alternative explanation, the metastatic theory, proposes that thyroglossal duct cyst carcinoma originates from an undetected primary thyroid tumor, given the multifocal nature of papillary carcinoma. [10] While Crile suggested the thyroglossal duct could serve as a pathway for thyroid cancer spread, this metastatic theory is generally considered less probable. [11] Mobini et al. argue that squamous cell carcinoma is likely the only true carcinoma arising directly from the thyroglossal duct, as other malignancies develop within ectopic thyroid tissue. Consequently, squamous cell carcinoma stands as the only primary thyroglossal duct cyst tumor, a rare entity with a poor prognosis and a mortality rate of 30–40%. [12]

Conclusion

The occurrence of carcinoma within a thyroglossal duct cyst (TGDC) is exceedingly rare, with papillary thyroid carcinoma being the most frequently reported histological subtype. Malignancy is often not suspected preoperatively, as the clinical and radiological presentation typically mimics that of a benign cyst. In most cases, the diagnosis is made incidentally during histopathological examination following surgical excision. However, the presence of certain imaging features—such as solid components, calcifications, irregular margins, or increased vascularity—should raise suspicion for malignancy. In such cases, surgical excision via the Sistrunk procedure remains the treatment of choice. Comprehensive gross examination and extensive histopathological sampling, particularly of any solid or atypical areas within the cyst, are strongly recommended to ensure accurate diagnosis and to rule out the presence of carcinoma. Given the potential for coexisting thyroid pathology, preoperative evaluation should also include ultrasound of the thyroid gland to assess for synchronous lesions, as well as fine-needle aspiration cytology (FNAC) in selected cases with high-risk features.

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