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Case Report

Incidental papillary carcinoma originating from a thyroglossal duct cyst: Diagnosis by histopathology & immunohistochemistry

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Abstract

Papillary thyroid carcinoma (PTC) arising from a thyroglossal duct cyst (TDC) is a rare, incidental diagnosis, occurring in less than 1% of TDC cases. These cysts typically present as asymptomatic midline neck swellings and are often not associated with any significant clinical symptoms or changes in laboratory parameters. A 28-year-old woman presented to the ENT outpatient department with a 13-year history of a slow-growing swelling on the left side of her chin and neck. The swelling was not accompanied by symptoms such as difficulty swallowing (dysphagia), difficulty breathing (dyspnea), or changes in voice. Comprehensive clinical evaluation, imaging studies, and laboratory analyses were conducted to assess and manage the case of PTC arising within the TDC. Current evidence suggests that performing a routine thyroidectomy may not significantly alter the prognosis of papillary carcinoma within a TDC, underscoring the need for a tailored approach based on individual patient assessments and risk factors.

Keywords: Thyroglossal duct cyst, Papillary thyroid carcinoma.

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1. Introduction

Thyroglossal duct cysts (TDC) are uncommon and typically present as midline neck swellings. They represent the most common congenital thyroid developmental anomaly during childhood, though diagnoses in adults are rare. Malignancy within a TDC is extremely rare, occurring in less than 1% of cases. TDCs account for about 70% of congenital midline neck masses in children, with only approximately 1% of these cysts associated with thyroid carcinoma. Papillary carcinoma arising from a TDC is an unusual and incidental finding, occurring in about 1% of TDC cases and showing a female-to-male ratio of 2.3:1.

2. Case Presentation

A 28-year-old female patient, came to the ENT outpatient department with history of swelling over the left side of her chin and neck for since 13 years with a slow-growing history. This swelling was not accompanied by symptoms such as difficulty swallowing (dysphagia), difficulty breathing (dyspnea), or changes in voice. There was no history suggestive of hypo or hyperthyroidism. There was no other significant history past medical history or relevant outcomes. (**Figure 1**).

On clinical examination irregular, tender, hard swelling of size measuring 10.0x 8.0 cm with multiple sinus tracts was present over midline neck region, mobile with deglutination. Routine thyroid function blood tests and investigations were in the normal range. MRI of neck suggestive of chronic benign cystic lesion (TBSRTC category II). FNAC was suggestive of a benign cystic lesion. The patient underwent Sistrunk's operation with complete removal of the thyroglossal tract and part of the hyoid bone between the lesser cornua along the tract. Histopathological examination revealed papillary thyroid carcinoma arising from TDC. IHC showed positivity for BRAF and thyroglobulin.

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Figure 1: Clinical image of the lesion showing sinus tract (lateral view)

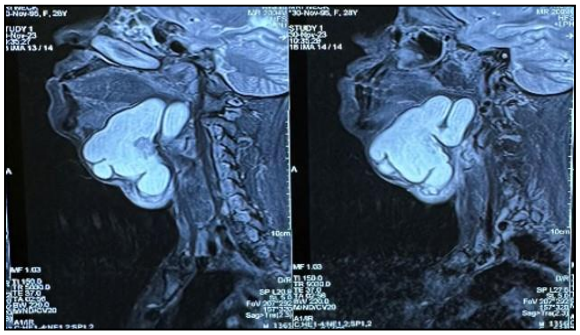

Figure 2: Non contrast MRI (Axial and Sagittal): Left loculated cystic lesion, likely benign cyst. (?Lymphoepithelial cyst

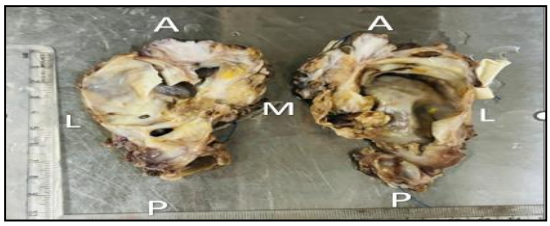

Figure 3: A cyst with body of hyoid bone measuring 8.5 x 6.0 x 3.5 cm. On cut section, 1cc straw coloured fluid expelled and multiple cystic areas with anterior surface showing skin and papillary excrescence. A: Anterior (skin), L: Lateral (cystic lining), M: Medial (hyoid bone), P: Posterior (cystic lining)

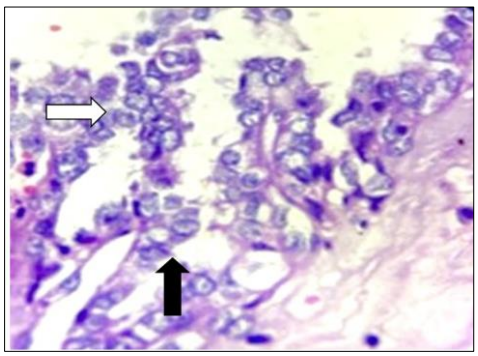

Figure 4: Individual follicular cell, shows overlapping (white arrow), & nuclear clearing (black arrow) (100X, H&E)

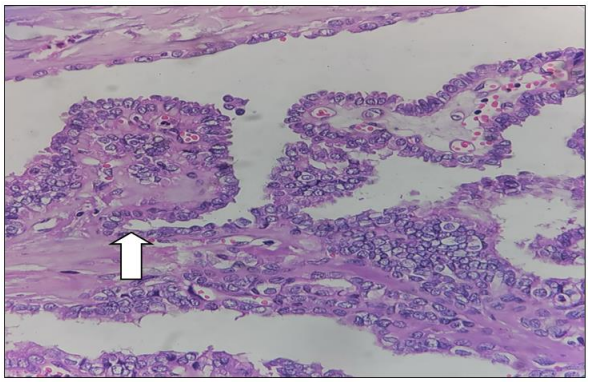

Figure 5: Follicular cells are arranged in papillary pattern (white arrow) with fibrovascular core (40x, HPE)

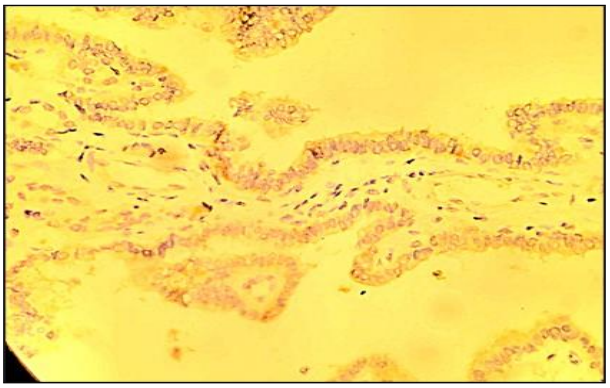

Figure 6: **BRAF**-mildly cytoplasmic positivity for anterior margin

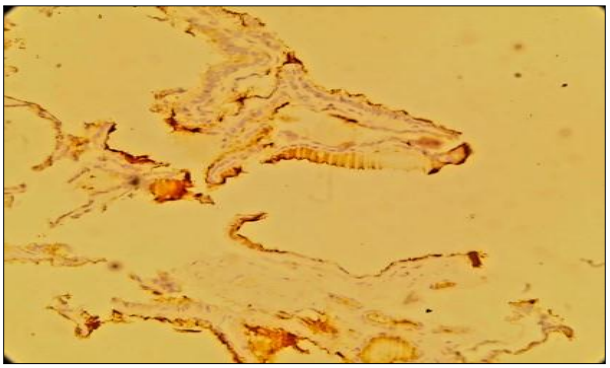

Figure 7: **Thyroglobulin (TG):** cytoplasmic positivity for medial margin

3. Discussion

Papillary thyroid carcinoma (PTC) originating from a thyroglossal duct cyst (TDC) is an unexpected and rare finding, representing less than 1% of TDC cases. Typically, TDCs present as benign midline cystic lesions without noticeable symptoms or abnormal clinical markers, making diagnosis challenging.[1]

Thyroglossal duct cyst carcinomas are more frequently diagnosed in females, generally between the third and sixth decades of life.[2] Papillary carcinoma is the most common type found in these cases, although mixed papillary/follicular carcinomas (approximately 8%) and squamous cell

carcinomas (around 6%) have been reported. Other rarer forms, such as Hurthle cell, follicular, and anaplastic carcinomas, account for an additional 6% of cases.[1,4]

Patients with TDC typically present with a painless, midline neck mass that moves upward when the tongue is protruded.[3,7] In rare instances, the cyst may appear on the floor of the mouth. While histopathological examination remains the definitive method for diagnosing TDC malignancies, fine-needle aspiration cytology (FNAC), ultrasound, computed tomography (CT), and immunohistochemistry (IHC) play crucial roles in differential diagnosis and confirmation of malignancy.[8]

Histologically, papillary carcinoma in a TDC is characterized by columnar cells arranged in papillae or follicular structures supported by a vascularized connective tissue stroma. These cells display distinctive features, including ovoid, ground-glass nuclei with grooves and pseudoinclusions, known as "Orphan Annie eye" nuclei. Psammoma bodies may also be observed.[4,5,7]

In this case, histological examination studied from the tumour proper shows follicular cells arranged in a papillary pattern with fibrovascular cores and branching patterns. The individual cells exhibited overlapping, nuclear clearing, and grooves, accompanied by psammomatous calcifications and sections studied from anterior margin shows skin with epidermis and dermis, subepithelium consists of chronic inflammatory cell infiltrates. Deeper dermis shows granulation tissue along with tumour proper located at distance of about 0.5cm from epidermis. Whereas lateral and posterior margins are negative for tumour while Medial margin consists of osteocytes and lacunae along with fibrocollagenous tissue along with tumour deposits as mentioned above. To confirm these margins, we have sent for IHC analysis for confirmation. No palpable lymphnodes where detected during the surgery, and after surgery follow up was done after 15 days and referred patient to higher centre due to involvement of margins.

IHC analysis is used to evaluate surgical margins, with BRAF showing mild cytoplasmic positivity at the anterior margin, and thyroglobulin (TG) exhibiting cytoplasmic positivity at the medial margin, confirming the thyroid origin.

The exact cause of TDC carcinomas remains unclear. Hypotheses include metastasis from an undetected primary thyroid tumour or the spontaneous development of carcinoma within ectopic thyroid tissue in the TDC wall.[1,6]

4. Conclusion

Malignancy within a TDC is rare and may be missed during radiological or FNAC evaluations. It should be considered in the differential diagnosis of a midline neck mass. The prognosis for TDC carcinoma is generally favorable, highlighting the importance of prompt and accurate histopathological diagnosis and IHC to guide effective treatment and improve patient outcomes.

5. Source of Funding

None.

6. Conflict of Interest

None.

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