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Papillary Carcinoma of Thyroid Arising in a Thyroglossal Duct Cyst

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ABSTRACT

Introduction: Thyroglossal duct cyst (TGDC) is the most common congenital midline neck anomaly. Malignant transformation is rare, occurring in approximately 1% of TGDCs, with papillary thyroid carcinoma (PTC) being the predominant malignancy. Preoperative diagnosis is challenging because malignant TGDC may clinically and radiologically mimic a benign cyst.

Materials and methods: A rare case of PTC arising within a TGDC was evaluated using clinical, radiological, gross, histopathological, and immunohistochemical findings. The relevant literature was reviewed to assess the diagnostic and therapeutic significance of this entity.

Case report: A 33-year-old woman presented with a painless, gradually enlarging midline suprahyoid neck swelling for four years. Ultrasonography demonstrated a well-defined cystic lesion containing solid nodules and multiple calcifications, with cervical lymphadenopathy. Contrast-enhanced CT suggested a TGDC with possible papillary carcinoma arising within it. Excision of the cyst was performed. Histopathological examination showed a cyst lined by flattened epithelium with focal thyroid tissue displaying papillary architecture, nuclear clearing, and psammoma bodies, diagnostic of PTC. Immunohistochemistry showed TTF-1 positivity, while BRAF V600 was negative. The findings supported PTC arising within a TGDC.

Conclusion: PTC arising in a TGDC is a rare entity that may remain clinically unsuspected. Careful histopathological examination is essential for diagnosis, while clinical, radiological and immunohistochemical correlation aids in establishing the diagnosis and guiding appropriate management.

Keywords: Thyroglossal duct cyst; Papillary thyroid carcinoma; Ectopic thyroid tissue; Psammoma bodies; TTF-1; Neck swelling.

INTRODUCTION

Thyroglossal duct cyst (TGDC) is the most common congenital developmental anomaly of the neck and results from persistence of the embryological thyroglossal duct during migration of the thyroid gland from the

foramen caecum to its definitive pretracheal position. It typically presents as a painless, midline neck swelling that characteristically moves with deglutition and protrusion of the tongue. [1,2]

Malignant transformation within a thyroglossal duct cyst is uncommon, occurring in approximately 1% of cases. Papillary thyroid carcinoma (PTC) is by far the most frequent malignant neoplasm arising in a TGDC, accounting for approximately 80–90% of reported malignancies. [2,4,6] Other malignant tumors, including squamous cell carcinoma, are considerably less common. The diagnosis is frequently unsuspected clinically because the presentation may closely mimic that of an uncomplicated benign thyroglossal duct cyst. [3,5]

Papillary carcinoma arising in a TGDC is thought to develop from ectopic thyroid tissue present within the cyst wall. Histopathological examination is therefore essential for establishing the diagnosis and demonstrating the characteristic nuclear features of PTC in association with thyroglossal duct remnants and/or ectopic thyroid tissue. [2,3]

We report a rare case of papillary thyroid carcinoma arising in a thyroglossal duct cyst in a 33-year-old woman, highlighting its clinical, radiological, histopathological and immunohistochemical features.

MATERIALS & METHODS

The present study describes a case of papillary thyroid carcinoma arising within a thyroglossal duct cyst in a 33-year-old woman. Relevant clinical findings, radiological investigations, and histopathological findings were reviewed.

The excised specimen was received in the Department of Pathology and subjected to routine gross examination. Representative tissue sections were processed, paraffin embedded and stained with hematoxylin and eosin (H&E) for microscopic evaluation.

Histopathological examination was performed to identify the morphological features diagnostic of papillary thyroid carcinoma. Immunohistochemical evaluation was performed using thyroid transcription factor-1 (TTF-1) and BRAF V600 immunohistochemistry. The final diagnosis was established by correlating the clinical, radiological, histopathological and immunohistochemical findings.

RESULT

CLINICAL FINDINGS

A 33-year-old woman presented with a painless swelling in the anterior midline of the neck since last four years. The swelling had gradually increased in size without associated pain, dysphagia, dysphonia, fever, or weight loss. There was no history of any discharge from the swelling.

On clinical examination, a 2x2 cm firm, non-tender anterior midline neck swelling was palpated in the suprahyoid region. The swelling moved upwards with deglutition and protrusion of the tongue. Bilateral cervical lymphadenopathy was palpated in level IB and level II.

RADIOLOGICAL FINDINGS

Ultrasonography revealed a 19x12 mm well-defined cystic SOL at midline of neck at suprahyoid location with solid nodules within and multiple calcifications, largest one measuring 1.5x0.89 cm without any internal vascularity on colour doppler study, likely suggesting Branchial cleft cyst. Both lobes of thyroid are normal in size and echotexture. Multiple enlarged lymph nodes were present in both lobes of thyroid at level IB & II, largest one measuring 17x8 mm at right lobe and 18x10 mm at left lobe.

CECT Scan of neck revealed features suspicious for-

i) Thyroglossal duct cyst (favoured, possibility of papillary carcinoma arising within).

ii) Complicated/infected thyroglossal duct cyst.

iii) Less likely cystic metastatic nodal disease from occult Papillary Thyroid Carcinoma.

Thyroglossal cyst excision was performed, and the specimen was submitted for histopathological examination.

HISTOPATHOLOGICAL FINDINGS

Gross Examination

One irregular tissue piece measuring 4x3x2 cm was received, calcified areas were also present.

Microscopic Examination

Sections showed histology of fibro-connective tissue with a small cyst lined by single layer of flattened cells. There is focal presence of thyroid tissue with papillary arrangement, clearing of nucleus and presence of calcified psammoma bodies. The histology was suggestive of papillary carcinoma of thyroid, involving the supplied tissue.

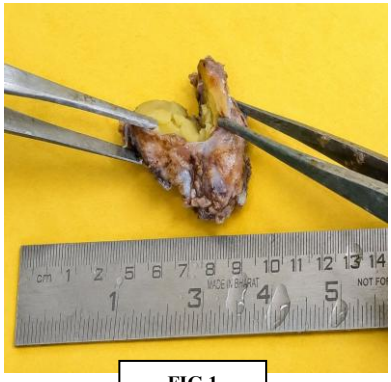
IMMUNOHISTOCHEMISTRY

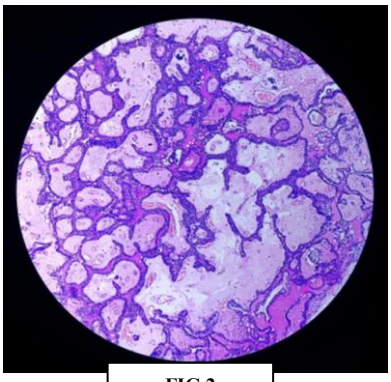
Immunohistochemistry reports were as follows:

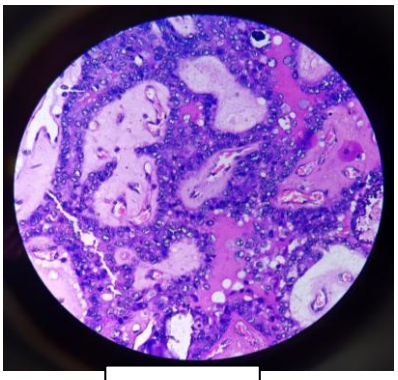
TTF-1: Positive

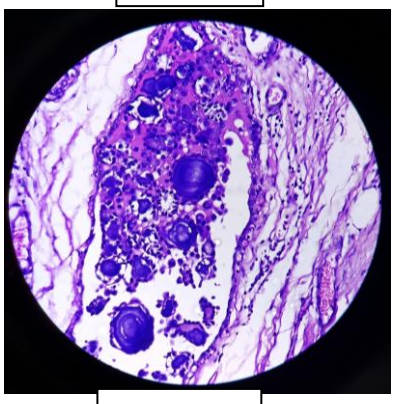
BRAF V600: Negative

Impression: IHC panel favours Papillary Carcinoma of thyroid.









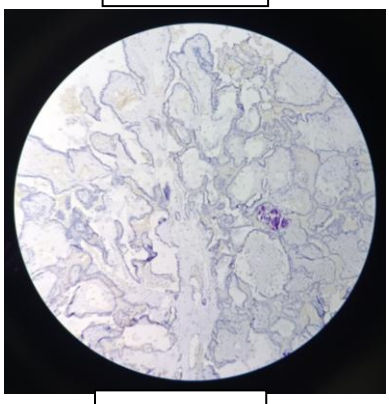


FIGURE 1: Gross picture of thyroglossal duct cyst with areas of calcification.

FIGURE 2: H&E(100X) Micrograph showing focal presence of thyroid tissue with papillary arrangement.

FIGURE 3: H&E(400X) Micrograph showing focal presence of thyroid tissue with papillary arrangement and clearing of nucleus.

FIGURE 4: H&E(400X) Micrograph showing calcified psammoma bodies.

FIGURE 5: Micrograph showing negative staining in neoplastic cells (100X monoclonal antibody against BRAF V600).

FIGURE 6: Micrograph showing positive staining in neoplastic cells (100X monoclonal antibody against TTF-1).

DISCUSSION

Papillary carcinoma of thyroid arising in a thyroglossal duct cyst (TGDC) is an uncommon entity, accounting for approximately 1% of all thyroglossal duct cysts. The vast majority of these malignancies are papillary thyroid carcinomas, while squamous cell carcinoma

and other histological subtypes are exceedingly rare. The tumour is believed to arise de novo from ectopic thyroid tissue within the cyst wall rather than representing metastatic spread from the thyroid gland, although synchronous thyroid carcinoma has been reported in 20–30% of cases. [1,6,7] Clinically, malignant TGDC is difficult to distinguish from a benign cyst, as most patients present with a painless, slowly enlarging midline neck swelling. Features such as rapid enlargement, fixation, cervical lymphadenopathy, or a hard mass may raise suspicion but are not consistently present. Consequently, the diagnosis is often established only after histopathological examination of the excised specimen.[1,7] Histopathological evaluation remains the gold standard for diagnosis. Typical findings include the characteristic nuclear features of papillary thyroid carcinoma arising within the wall of a thyroglossal duct cyst, with evidence of normal thyroid tissue in the cyst wall supporting a primary origin. Current management is guided by risk stratification. The Sistrunk procedure is considered adequate treatment for low-risk patients with disease confined to the cyst, whereas total thyroidectomy, with or without radioactive iodine therapy, is recommended in selected high-risk patients, including those with thyroid gland involvement, lymph node metastasis, tumour size >1 cm, extrathyroidal extension, or adverse histological features.[6,8,9] Overall, the prognosis is excellent, with reported long-term survival rates exceeding 95%, reflecting the indolent biological behaviour of papillary thyroid carcinoma arising in a TGDC when appropriately managed.[7,8]

CONCLUSION

Papillary carcinoma of thyroid arising in a thyroglossal duct cyst is a rare clinicopathological entity that is often diagnosed only after histopathological examination of an excised TGDC. Though histopathology remains the gold standard for diagnosis of papillary carcinoma of thyroid, a proper clinical, radiological and immunohistochemical correlation can help in suspecting as well as early diagnosis allowing early intervention in view of poor prognosis of the lesion. Awareness of this rare entity is important for both surgeons and pathologists to ensure appropriate diagnosis and management.

Declaration by Authors

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