



Case Report

PAPILLARY CARCINOMA IN THYROGLOSSAL DUCT CYST – A RARE CASE REPORT

Article History:

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Received: 20-02-2026

Revised: 27-02-2026

Accepted: 18-03-2026

Published: 22-03-2026

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INTRODUCTION

Thyroglossal duct cyst is the most common congenital thyroid gland disease accounting for over

Abstract: INTRODUCTION : Thyroglossal duct cysts are usually presented as swelling in the midline of neck. Co-existence of carcinoma in the cyst is very rare to find. Diagnosis can be made only after excision of cyst in post operative period. Sistrunk operation is usually considered adequate but the need for thyroidectomy depends entirely on histopathology findings. We are here reporting a case of 24 year old female diagnosed to have papillary carcinoma within the thyroglossal duct cyst. **CASE REPORT:** A 24 year old female presented with midline neck swelling for 4 months. She had no history of hypo or hyperthyroidism symptoms and no compression symptoms either. CT neck revealed well defined bilobed cystic lesion of 4*1.7 cms with eccentric mural nodule of 8*6.7mm. Thyroid function tests were within normal limits. FNAC revealed submental swelling suggestive of benign cystic lesion. Underwent Sistrunk procedure under general anesthesia. Revealed as papillary carcinoma thyroid on HPR. **DISCUSSION:** Thyroglossal duct carcinoma usually presents in 4th decade of life and more prevalent in women than men. It is embryologically arising from the remnants of thyroid gland. Over 90% of cases are noted to be papillary in origin. As initial workup ultrasound neck and CT may be done to confirm the presence of thyroid gland. FNAC adds further information however HPR gives us the confirmatory diagnosis only in post operative period. **CONCLUSION:** In low-risk patients, Sistrunk operation may be enough whereas in high-risk patients further more aggressive modality of treatment may be needed to make the patient completely disease-free.

Keywords: Papillary thyroid carcinoma, Thyroglossal duct cyst, Sistrunk procedure, Thyroidectomy.

two-third cases of midline neck swelling in children. Out of those only 1% have the risk of transforming into a carcinoma. The first ever case was reported by Brentano in 1911⁽¹⁾.

Thyroid gland descends from foramen caecum to its location at a point below the thyroid cartilage. The epithelial tract left behind is called the thyroglossal tract. The development of thyroid gland begins at 3rd week of gestation and this tract usually gets disappeared by 8 to 10 weeks of gestation. Failure of this tract to close or incomplete atrophy of this thyroglossal tract forms the origin for thyroglossal duct cyst. Thyroglossal remnant may be a cyst, tract, duct, fistula or any ectopic thyroid gland within a cyst.

Definitive treatment is the excision of cyst along with removal of entire tract with branches. Any relation with hyoid bone may lead to removal of central part of hyoid bone to ensure complete removal. Recurrence is rarely noticed except in case of intraoperative cyst rupture. In patients who are diagnosed with papillary carcinoma of thyroid, thyroidectomy is preferred in those who had enlarged lymph nodes, h/o neck irradiation and cold nodule with I₂ uptake.

RESULTS

A 26 year old female presented with chief complaints of swelling over anterior aspect of neck for about 4 months, associated with pain since 2 weeks. There are no other hypothyroidism or hyperthyroidism symptoms. There are no compression symptoms like dysphagia or hoarseness of voice. No prior radiation exposure. No significant family history noted.

Examination Findings: On Inspection there is a single ovoid swelling of 3*2 cms over sub mental region extending 2cms below symphysis menti and 2cms above the hyoid cartilage. The swelling is moving with protrusion of tongue and deglutition. Skin over swelling is normal and surface is smooth with no scar/ discharge/ sinus or discolouration. Trachea appeared midline.

On Palpation: Inspectory findings of 3*2 cms ovoid swelling in submental region confirmed. Soft to cystic in consistency. Skin over the swelling is pinchable. No local rise of temperature. Swelling is freely mobile from side to side. Plane of swelling is in deep to deep fascia. Fluctuation present. Mild tenderness present. Non compressible. Non pulsatile. Trachea in midline. Bilateral carotids felt. There is no evidence of any lymph nodal enlargement.

On Investigation: CT NECK - A well defined bilobed cystic lesion measuring 4*1.7cms with an eccentric enhancing mural nodule measuring 8*6.7mm noted in midline towards right side at the level of hyoid and sub hyoid region around the right genohyoid attachment site. Lesion separate from submandibular gland. Few bilateral level II cervical lymph nodes noted with maintained fatty hilum, largest measuring 10*5mm on right and 11*7mm on left side. Thyroid function test is within normal limits. FNAC of submental swelling – features suggestive of BENIGN CYSTIC LESION.



Figure 1: Image of CT NECK showing Bilobed cystic lesion with few enhancing lymph nodes.

PROCEDURE: Underwent Sistrunk Procedure under general anesthesia. **Histopathology impression:** CLASSICAL PAPILLARY CARCINOMA THYROID.



Figure 2 : Specimen of thyroglossal duct cyst .

IHC markers: GALACTIN -positive in tumour cells. CK -19 diffuse and strong positive in tumour cells. Impression: PAPILLARY CARCINOMA THYROID - CLASSICAL TYPE.

PET CT- Post excision status of cystic lesion in submental region. Mild metabolically active fluid collection in submental region with air locules- ?post operative seroma. No other abnormal metabolically active disease in rest of the regions of body.

Oncology opinion obtained and advised to follow up as there is no metabolically active disease.

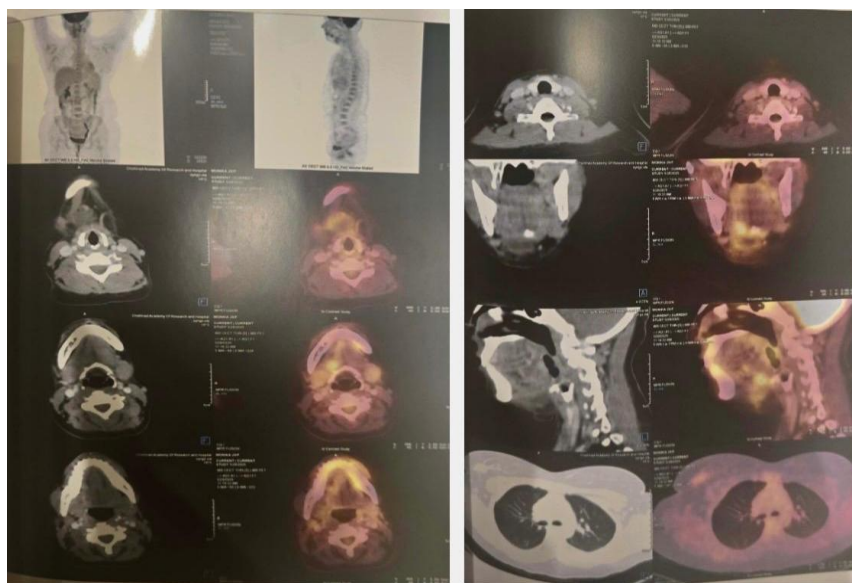


Figure 3: Preoperative 18F-FDG PET/CT showing a midline thyroglossal duct cystic lesion with a focal FDG-

avid mural nodule, raising suspicion of papillary carcinoma. Whole-body maximum intensity projection and selected axial, coronal, and sagittal CT, PET, and fused PET/CT images are illustrated.

DISCUSSION

Thyroglossal duct cyst carcinoma is the one of the rare entity and mostly seen in 4th decade of life with female preponderance in the ration of 3:2. TGDCC can be presented as asymptomatic mass in the neck or as a rapidly growing mass with tenderness.² Risk of transformation into malignancy is upto 1% for TGDC.³ Histologically upto 90% are papillary carcinoma and they are multicentric and multifocal growth is noted as about 10% cases present with second malignant lesion in thyroid cancer.⁹ Upto 5% are squamous in origin and other types include Hurthle cell carcinoma, anaplastic and follicular cell carcinoma.⁴

Two possible theories explain the etiology. First is the DENOVO THEORY and the most acceptable one as histopathology revealed ectopic thyroid tissue in 64% of cases and it is further approved by absence of medullary variant of thyroid carcinoma which arises from the para follicular cells.⁵ Second theory is the METASTATIC THEORY stating its origin from an occult thyroid gland.⁶ Mobini.et.al considered squamous cell carcinoma to be the only true carcinoma of thyroglossal duct as others arise from ectopic thyroid tissue.⁷ Squamous cell carcinoma is extremely rare to find and prognosis is poor with mortality rate upto 35%.⁸

Neck ultrasound is done as initial radiological workup to evaluate the cyst. Ultrasound features of TGDCC are calcifications, presence of mural nodule or solid component and regional lymphadenopathy.¹⁰ Next to that , CT imaging or MRI may be done to confirm ultrasound findings and identify calcifications, solid nodules , vascularity and margin irregularity.¹¹

Diagnostic potential of FNAC is controversial. It can detect malignancy in upto 53% and even more effective if solid component is sampled. In our case, cytology is suggestive of benign lesion.

No proper guidelines were laid on the management part but Sistrunk procedure is being carried out as initial treatment and it is offering a cure rate upto 95% in papillary carcinoma of TGDC with excellent prognosis of 95-100% unlike squamous type.^{R Patel et al} did not find any significant improvement in outcome of patients who underwent total thyroidectomy. Few other authors also emphasized that the extent of initial surgery was the sole predictor of overall survival outcome. Moreover complications like hypocalcemia and recurrent laryngeal nerve injury may happen with total thyroidectomy.

It was suggested to proceed with Sistrunk surgery

alone low risk individuals : age <45 years, tumour sized <1-1.5cms, radiologically normal thyroid gland, no h/o prior irradiation, presence of negative margins and absence of metastatic lymph nodal spread. In high risk patients total thyroidectomy may be preferred as a followup surgery. As per recent analysis SP, TT , Neck dissection and RAI are recommended as ideal approach in patients over 45 years age, tumour size >4cms, with surrounding soft tissue involvement, cold nodule in thyroid gland , lymph nodal or distant metastasis.

Follow up is advised with postoperative RAI, suppressive dose of levothyroxine therapy and a whole body scintigraphy for best outcomes. Multidisciplinary collaboration and further case studies may relieve the controversies in management part of TGDCC.

CONCLUSION

Thyroglossal duct cyst carcinoma is always a post operative histopathology confirmatory diagnosis that may be missed due to its rarity. Coming to the management of thyroglossal duct cyst carcinoma, in low-risk individuals with no positive tumour margins on HPR, simple Sistrunk procedure is enough. But if at all patients belong to high risk group more aggressive approach may be needed to completely resolve the pathology. Thyroidectomy was advised in cases with enlarged lymph nodes, h/o neck irradiation and cold nodule with I2 uptake. Further case survey may resolve the controversies regarding management options.

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