

## A Rare case of papillary carcinoma in branchial cleft cyst

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Anomalies of branchial cysts are rare diseases of the head and neck region. Confirmation of definite pathology is always challenging for clinicians. Ectopic thyroid tissue is ubiquitous in the body and a branchial cleft cyst a congenital abnormality can rarely harbor thyroid tissue in the cyst. Primary papillary thyroid carcinoma (PTC) inside ectopic thyroid tissue is extremely rare. We report a very rare case of PTC incidentally arising in a branchial cyst cleft.

**Keywords:** Branchial cleft cyst, Papillary carcinoma, Thyroid

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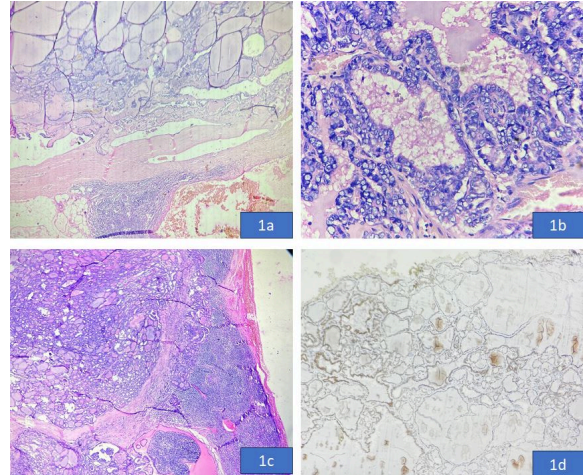
## Introduction

Branchial cleft cysts are commonly encountered in lateral neck swelling in clinical practice, however, the presence of ectopic thyroid tissue with papillary thyroid carcinoma (PTC) is extremely rare [1]. PTC primarily arising in branchial cleft cyst is indistinguishable from metastatic papillary carcinoma in the cyst and the diagnosis can only be confirmed after total thyroidectomy to exclude occult primary [2]. The first case of PTC in ectopic thyroid of the branchial cyst was reported in 1992 by Balasubramanniam et al [3]. We hereby report a very rare case of papillary thyroid carcinoma incidentally arising in a branchial cleft cyst. To the best of our knowledge, only seventeen cases have reported PTC arising in a branchial cyst without a primary in the thyroid [3], [4], [5], [6].

## Case Report

A 70-year-old male presented with a painless right-sided lateral neck mass. Physical examination revealed a 4 cm mass on right side of neck. No significant cervical lymphadenopathy or thyroid mass was noted. Contrast-enhanced computed tomography (CECT) of neck showed a well-demarcated walled, solid cystic-enhancing mass lesion in right carotid space of neck. The solid component was measuring 2.6 mm in size and showed peripheral foci of calcifications. No cervical lymphadenopathy was detected. The differential diagnosis offered in radiology were vagal schwannoma, nerve sheath tumor and paraganglioma. The patient underwent surgical excision of mass and histopathological examination revealed a 5 cm cystic mass with keratin debris confirming a branchial cleft cyst. A solid area measuring 2 x 1.5 cm was identified. Microscopic examination revealed a cyst wall lined by cuboidal epithelium and stratified squamous epithelium with a fibrotic wall showing dense lymphoid infiltrate. The wall also showed few foci of normal thyroid tissue and classical type papillary carcinoma comprising of tumour arranged in form of papillae lined by cells showing nuclear overcrowding, grooving and intranuclear inclusions. Psammoma bodies were also identified. No areas of necrosis were identified. On immunohistochemistry, tumour cells were positive for TTF-1, PAX-8, p-16 and were immunonegative for p63. Two out of four lymph nodes in wall showed metastatic deposits of papillary carcinoma.

To rule out primary thyroid carcinoma and any residual malignancy, a Tc-99m Sestamibi scan was performed, which did not reveal any increased uptake in the thyroid gland. The patient has been under regular follow-up for 6 months, there is no evidence of recurrence.



**Figure: 1 H&E-stained sections, x40, showing branchial cyst wall and tumor. H & E stained section, 400x, showing papillary architecture of tumor cells H & E stained section, 40x, showing lymph node metastasis IHC, TTF1 positivity**

## Discussion

The thyroid is the first endocrine organ to develop during fetal embryology and is derived from endoderm. Failure of the thyroid to descend from the thyroid anlage region to its final location in front of the trachea can give rise to ectopic thyroid tissue. The most common location of ectopic thyroid is in the midline-lingual, high cervical, trachea, superior mediastinal and even pericardiac. Very rarely, it can be located in the lateral cervical region. One of the most common cystic lesions of the lateral head-neck region is a branchial cyst. These are located anteriorly to the sternocleidomastoid, although some rare sites have also been reported. The etiology of branchial cysts is not clear, various theories suggest congenital malformation, and cystic degeneration of lymph nodes due to epithelial migration [7]. The proximity of branchial arches during development can explain the presence of ectopic thyroid tissue. The literature studies reveal that ectopic thyroid tissue may clinically present as primary thyroid carcinoma especially papillary carcinoma,

However, most of these reports are from thyroglossal duct cysts, lingual thyroid and struma ovarii. Primary papillary carcinoma of the thyroid arising in branchial cysts is rare. Cochrane search shows only 17 cases have been reported in the literature as PTC arising from branchial cysts without a primary in the thyroid [6]. Two of these cases presented with metastatic lymph node disease at diagnosis. The usual diagnostic dilemma in such situations is that carcinoma in the cyst is a primary or metastatic disease with missed/occult primary in the thyroid. Sidhu et al. proposed the following criteria for diagnosing papillary thyroid carcinoma in branchial cysts [8].

1. an epithelial lining layer with subepithelial lymphoid tissue collection,
2. normal thyroid tissue adjacent to the focus of papillary carcinoma within the wall,
3. and no evidence of papillary carcinoma in the thyroid or other areas.

In our case, tumor showed morphological features of papillary thyroid carcinoma and fulfilled all the above criteria. Metastases in neck nodes are detected in about 20% of cases as per the available evidence and distant metastasis has not been reported [9], [10]. Immunohistochemical positive staining for TTF-1, PAX-8 and thyroglobulin can help in the identification of a PTC, however, it cannot distinguish between primary and metastatic tumors. CT, MRI and scintigraphy imaging can help in detecting ectopic thyroid tissue, however histopathological examination is the gold standard.

## Conclusion

In conclusion, we have reported a very rare case of primary PTC located in the branchial cyst. Clinicians should always keep the possibility of ectopic thyroid tissue while dealing with the management of lateral neck mass. The diagnosis of this rare entity requires a multidisciplinary team comprising of radiologists, surgeons and pathologists.

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