

Case Report

Papillary carcinoma in a thyroglossal cyst: a rare case report and review of literature

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ABSTRACT

Thyroglossal cysts are the most common thyroid anomaly with a majority of it being diagnosed at fourth decade of life. Authors report a case of 32-year-old lady with a globular swelling of 3×2 cm moving with deglutition with no cervical lymph nodes and a normal thyroid function test. Intra operatively a distinct lesion of 3×2 cm was found superior to the thyroid gland with firm nodules in the right lobe of the thyroid. Patient underwent total thyroidectomy with central compartment neck dissection and Sistrunk's operation followed by a negative radio iodine scan and thyroid hormone suppression. This was considered based on intraoperative nodularity of the thyroid gland.

Keywords: Papillary carcinoma, Thyroglossal cyst, Total thyroidectomy Sistrunk's operation

INTRODUCTION

Thyroglossal cysts are the common anomalies in the embryological development of the thyroid gland. Thyroid gland develops from foramen cecum at the base of the tongue and descends below the thyroid cartilage leaving behind an epithelial tract which disappears at 8th to 10th week of gestation.

In adult, thyroglossal duct becomes obliterated whereas it remains as duct, cysts or fistula in 7% of population. Thyroglossal cysts can give rise to 1% of thyroid carcinoma.¹ Carcinomas occurring in thyroglossal cyst are extremely rare and only about 250 cases reported in literature.²

The median age of presentation is 40 years and most patients are females. Still controversies prevail regarding the need for thyroid gland removal and the post-operative follow up. Here, authors report a rare case thyroglossal cyst malignancy which mandated surgery.

CASE REPORT

A 32 year-old-lady presented with a swelling in front of the neck since four years. She neither had a history of thyroid malignancies in the family nor underwent any head and neck radiation. On examination, it was a globular swelling of 3×2 cm, just lateral to midline and about 3 cm below the hyoid bone. The swelling moves with deglutition but not with tongue protrusion. There were no other nodules were palpable in the thyroid and no palpable cervical lymphadenopathy. Thyroid function tests were within normal limits. Thyroid ultrasonography revealed heterogeneous and hypo echoic nodules of varying sizes in right lobe of the thyroid. Fine needle aspiration cytology (FNAC) from the lesion suggested Bethesda category 5 (suspicious of papillary carcinoma with a predominant follicular pattern).

Hence, authors proceeded to surgery and intra operatively authors noticed a distinct firm lesion of 3×2 cm separate from the thyroid gland and was found to be a thyroglossal cyst. Right lobe of the thyroid showed multiple firm

nodules measuring 1×1 cm. The patient underwent total thyroidectomy with central compartment neck dissection along with Sistrunk's procedure. Histopathology demonstrated papillary carcinoma with follicular predominance in the thyroglossal cyst with benign nodules in the right lobe of the thyroid (Figures 1 and 2). Postoperatively, the iodine-131 uptake scan revealed no activity in the remnant thyroid bed. The patient was commenced on thyroid hormone suppression therapy. At 6 months, the follow up radio iodine scan was unremarkable with no features of recurrence.

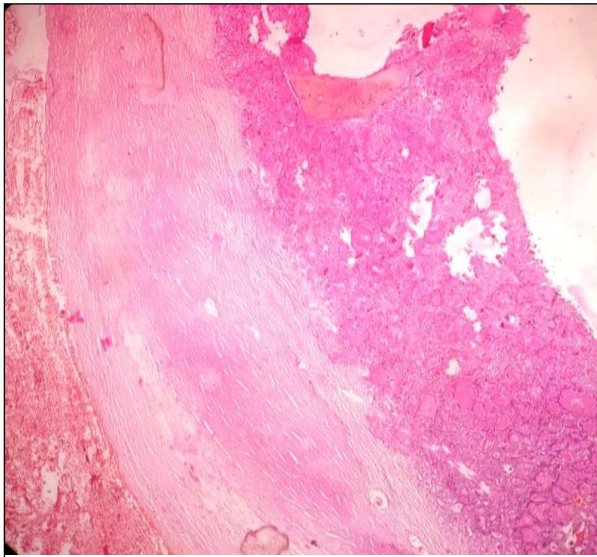


Figure 1: Section from the thyroglossal cyst shows cyst wall with tumour (H/E, 100X).

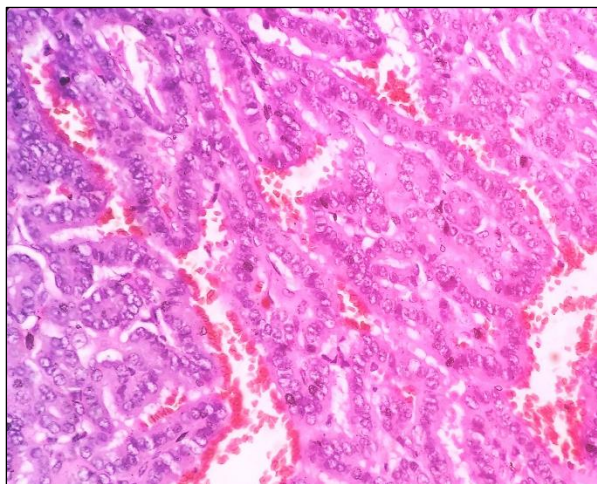


Figure 2: Section shows tumour follicles, lined by cuboidal cells having round nuclei with inconspicuous nucleoli (H/E, 400X).

DISCUSSION

Thyroglossal cyst is a frequent developmental anomaly during childhood. Some of the cases are identified in adults and accounts for about 7% of the population.

Carcinomas occurring in thyroglossal cyst are extremely rare, consisting of less than 1% cases. The most frequent histological type is papillary pattern in more than 80% followed by mixed variety, squamous cell, Hurthle cell, follicular and anaplastic variety.³

The etiology of carcinoma in a thyroglossal cyst is unknown and neither good clinical history nor an examination can lead to a preoperative diagnosis. The most accepted theory of the papillary carcinoma arising in a thyroglossal cyst is explained by de novo origin which postulates that carcinoma in the thyroglossal cyst arises from ectopic thyroid nests of the cyst wall. Second is the theory of metastatic spread from a primary thyroid tumor.⁴

Mostly carcinoma develops in the fourth decade of life and can occur in the any site in the embryological descent of the gland. Criteria for thyroglossal cyst carcinoma as proposed by Mesolella et al includes presence of a thyroglossal remnant, ectopic thyroid nests within the cyst wall and a clinically normal thyroid gland.⁵

Most of the cysts are benign, small in size and confined to the neck. The features suggestive of malignancy includes hard or fixed cyst with or without nodal involvement in 7% to 15% of the cases. Examination findings, imaging techniques (ultrasound, computed tomography, scintigraphy) cannot diagnose it preoperatively with FNAC having a very low sensitivity of 53%.^{5,6} Therefore carcinoma in the thyroglossal cyst is often a histopathological diagnosis.

Histopathologically, the major subtypes include papillary in 94% and squamous in 6%. Papillary has the best prognosis contrasting to squamous type having the worst prognosis. Merely a Sistrunk's surgery suffices for papillary carcinoma whereas Sistrunk's along with total thyroidectomy is recommended for squamous carcinoma of a thyroglossal cyst.⁷

Besides the difficulties in the diagnosis, there is lack of consensus regarding the surgical treatment of a thyroglossal cyst. Surgical approaches were suggested based on the view that papillary thyroid carcinoma may spread through the thyroglossal duct remnant even with no lesion detected clinically in the gland, leaving the patients be treated with Sistrunk's procedure and total thyroidectomy.⁸ Meticulous dissection is needed to avoid total thyroidectomy related hypocalcemia.⁹ Studies suggest total thyroidectomy if the cyst wall is invaded by the tumor or if the size of the tumor is greater than 1.5 cm. Certain authors favour thyroidectomy if the thyroid gland is found to be nodular, a cold nodule in a thyroid iodine uptake scan, clinically positive neck node and/or history of neck irradiation. In our case the tumor was more than 1.5 cm with no cyst wall invasion and a negative neck node status. Total thyroidectomy was done in view of tumour size and nodularity of right lobe.

Thyroid hormone suppression therapy is propagated irrespective of thyroid status.^{10,11}

CONCLUSION

Sistrunk's operation with a total thyroidectomy followed by thyroid hormone suppression after a negative radioiodine scan is done in this case after consideration of the embryological origin of the thyroglossal cyst and intraoperative nodularity of the thyroid gland. This concludes control of the disease and subsequent monitoring with thyroglobulin and radioiodine uptake study would be feasible for assessment of further recurrence. Due to rarity and vagueness of the presentation, thyroglossal cyst malignancy can be missed, causing a dilemma not only in its management, but also to that of the thyroid thus mandates aggressive evaluation.

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