



RARE COHABITATION OF PAPILLARY CARCINOMA IN THYROGLOSSAL DUCT CYST : A CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT **Introduction:** Papillary Carcinoma originating from thyroglossal duct cyst (TGDC) is very rare, occurring in less than one percent of cases. The diagnosis is often made in these situations following the removal of benign TGDC. We provide a case report that details a case of a thirty-years man who had Sistrunk's surgery for TGDC and was subsequently diagnosed to have Papillary Cancer. It is debatable how to handle these cases; the options include either a periodic monitoring following Sistrunk's Procedure or Complete thyroid gland excision +/- radio-active iodine (RAI) ablation and suppressive hormonal therapy. **Case Presentation:** A thirty-years man came to our OPD with anterior midline neck swelling which moved upward with deglutition and tongue protrusion. On radiology, distinct cystic lesion was seen. Sistrunk's Procedure was performed and tissue was sent for histo-pathological reporting. A papillary carcinoma foci was identified in the TGDC by histo-pathology. As a follow up and completion treatment, our patient was subjected to complete thyroidectomy. **Discussion:** The most frequent anomalies are TGDC which are encountered in thyroid development. Within TGDC remains, malignancy is present in one to two percent of cases with most being Papillary Carcinoma. TGDC are most common in females with ratio being 3:2.

KEYWORDS : Papillary Carcinoma, Thyroglossal Duct Cyst, Sistrunk's Procedure, Head & Neck Cancer

INTRODUCTION

A frequent congenital thyroid gland defect, TGDC account for seventy percent of midline neck masses in children and seven percent in adults¹. Approximately one percent of these cases occasionally give rise to carcinoma. First case was reported by Brentano followed by Uchermann in 1915. Only 300 cases have been documented in the literature till date^{2,3}.

We report a thirty-year-old male patient's case of papillary cancer that originated in a TGDC. Though the diagnosis of malignancy was missed on FNAC, papillary carcinoma incidentally occurred in TGDC on histopathology reporting after excision by Sistrunk's operation. The thyroid gland and cervical lymph node were unaffected. It is debatable how to manage these cases; the options include either a routine follow-up following Sistrunk's Procedure, or Total Thyroidectomy +/- radioactive iodine ablation and hormone therapy⁴.

CASE REPORT

A thirty-years man came to our OPD with anterior midline neck swelling noticeable since 6 month (Fig.1A). He did not exhibit compressive symptoms, hyperthyroidism, or hypothyroidism. Systemic examination revealed nothing noteworthy. He refuted any radiation exposure and the thyroid malignancy in his family history. On local examination he had a three by two cm oval, firm, non-tender swelling just lateral to midline of the neck. The swelling moved upward with deglutition and tongue protrusion. There were no palpable cervical lymph nodes or further thyroid gland nodules. The testing results for thyroid gland function were regular.

On USG Neck, a distinct anechoic cystic lesion beneath the hyoid bone, measuring 32 by 22 mm, without any solid peripheral component with an impression of thyroglossal duct cyst (TGDC) was seen (Fig.1B). FNAC revealed a benign cystic lesion. On MRI Scan there was a distinct cystic lesion with smooth, thin walls that measured 33 x 22 mm and was located in the midline, somewhat below the level of the hyoid bone (Fig.1C).

The patient had Sistrunk's surgery done. A longitudinal incision was taken across the mass, and the hyoid bone rim and a 32 mm TGDC that was inferior to it were both removed in whole (Fig. 2A, B, C). The postoperative phase went without incident. To our surprise final HPR revealed TGDC with accessory thyroid showing Papillary Thyroid Carcinoma (Fig.2D). Taking the patient's age into account, tumour size and histopathology report, total thyroidectomy was planned. The patient underwent total thyroidectomy, and was discharged after an

uncomplicated post-operative period. Thyroid function test and USG neck were advised to the patient on follow up along with Anti-Thyroglobulin Antibodies and Thyroglobulin levels. The Thyroglobulin levels were < 0.2 ng/ml, so patient was deemed to be low risk and was not subjected to Radio-active Iodine ablation.

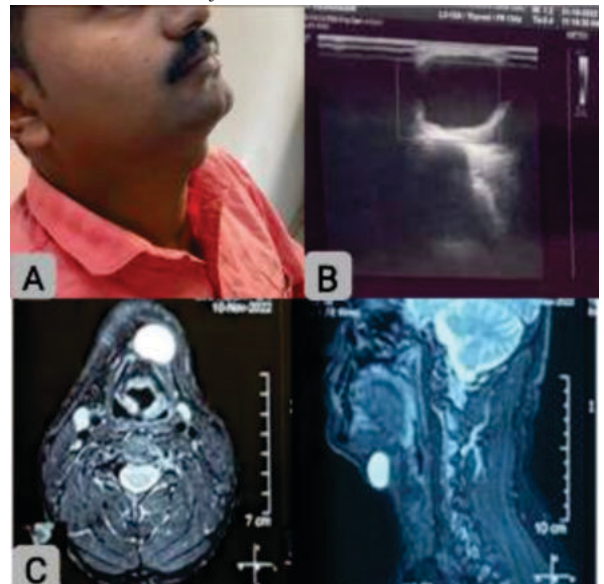


Fig.1. (a) Anterior Midline Neck Swelling (b) USG Showing Anechoic Cystic Lesion (c) MRI Showing Cystic Lesion Below Hyoid Bone in axial and sagittal section.

DISCUSSION

Thyroid anlage structures near the tongue's foramen cecum descend as a diverticulum during the third and fourth weeks of pregnancy, maturing into a region in front of the thyroid cartilage by the seventh week. We refer to this diverticulum as the thyroglossal duct. Regularly, this duct obliterates, yet on the off chance that it stays patent, it might shape as a cyst that may be found in any place from foramen cecum to the thyroid isthmus. The majority of midline neck masses in children are thyroglossal cysts⁵. Individuals aged 1-82 years might have TGDC. The fourth decade of life is when it is most prevalent, with an expanded pervasiveness in females than males (F:M, 3:2)².

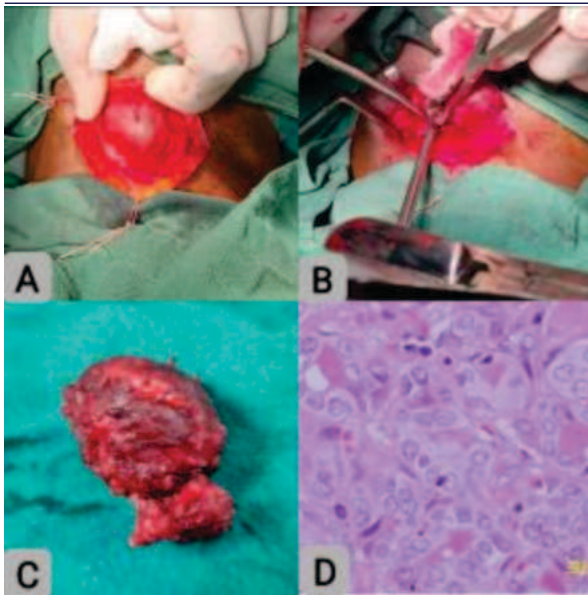


Fig.2. (a) Steps Of Sistrunk's Operation- CYST Wall Dissected Free (b) Body Of Hyoid Bone Resected Free (c) Thyroglossal Duct CYST Specimen With Hyoid Bone (d) Histopathology Showing Features Of TGDC With Papillary Carcinoma Thyroid.

In total, there is a one percent chance of malignancy in TGDC patients. Based on histology, the most often found malignant growth in TGDCs is papillary carcinoma, accounting for 80% of cases. Nonetheless, instances of follicular/mixed papillary cancer (eight percent) and SCC (six percent) have been accounted for. Anaplastic, follicular, and Hurthle cell carcinomas are three more intriguing and uncommon forms of carcinomas that make up six percent of cases. Papillary carcinoma was the condition that our patient had².

The etiology of TGDC carcinoma stays obscure; However, potential theories incorporate occult primary carcinoma metastasis or an unconstrained ectopic thyroid tissue evolution in wall of TGDC. The latter one most widely acknowledged. The fact that TGDCs do not have medullary carcinomas supports this conclusion.^(2,10) It is necessary to do TGDC examination in order to assess the cyst and verify the thyroid gland's existence. USG features suggestive of TGDC carcinoma incorporate regional lymph node enlargement, solid components and calcification. USG and CT of our patient uncovered no malignancy characteristics².

However, the location of TGDC carcinoma has a fifty three percent diagnosis probability, which might increment assuming a sample of the solid part is taken. The FNAC findings in this patient revealed a benign cystic lesion. Diagnosis of the thyroglossal cyst histologically (existence of the follicles of the thyroid in the cyst wall), no evidence of primary neoplasm in thyroid gland, the presence of normally functioning thyroid tissue neighbouring the malignancy are the three criteria for confirming papillary carcinoma in TGDC.

Considering that papillary carcinoma has the potential to disseminate through the thyroglossal duct remnants even in cases where there is no clinically distinguished disease in the gland, Sistrunk's procedure performed, combining a total thyroid removal and hormonal suppression is the optimal treatment⁶. Skip metastasis are more common in TGDC carcinoma. This brings the disease under command, a follow-up study using thyroglobulin and radio-iodine take-up might be conducted to evaluate any potential recurrences in years to come⁷. Post-operative monitoring is crucial, considering the elevated rate of concurrent thyroid gland neoplasms.

On the other hand, different authors pushed Total Thyroidectomy as a subsequent surgery in patients with a TGDC carcinoma because of engagement of thyroid in 33 to 45 percent instances prompting radioactive iodide (RAI) ablation and Thyroglobulin level is used as a monitoring indicator². Optimal follow-up strategy, according to experts, involves post-surgery RAI ablation, a suppressive dosage of L-thyroxine therapy and whole-body scintigraphy.

Ninety-eight publications on review showed that in 164 patients.

39.5(9-83) years was the average age; 68.3 percent of those examined were female. Definitive pathologic study revealed findings in 73.3 percent of cases overall. Papillary carcinoma accounted for the majority of pathologies (92.1 percent). Among the patients, Sixty-one percent had a complete thyroidectomy and 98.9 percent had Sistrunk's operation. The mean period from the start of therapy to the recurrence was 42.1 months, representing a 4.3 percent recurrence rate. At the time of the most recent follow-up, all of the other participants were disease-free, but one patient passed away with TGDC cancer⁸. With a survival percentage of 95.6 percent in a decade, papillary carcinoma emerging in TGDC has an outstanding prognosis⁵.

CONCLUSION

FNAC is an uncommon preoperative diagnostic tool in TGDC Carcinoma. In such cases, negative radioiodine scanning is performed. nodularity within the thyroid gland seen intra-operatively and the embryological basis of the TGDC must be taken into account. As an intriguing and rare disease, TGDC carcinoma management stays questionable. Malignancy in TGDC is frequently overlooked due to the disease's uncommonness and ambiguity, which makes it a challenge to manage both the thyroid and the cyst. This subsequently necessitates forceful assessment and it ought to be overseen in each instance with multifaceted strategy. Our case study adds to the body of knowledge on such an uncommon malignancy. To conclude TGDC carcinoma is a rare disease but carries excellent prognosis.

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