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Unifocal Papillary Carcinoma of the Thyroglossal Tract in A 25-Year-Old Female - An Uncommon Clinical Entity

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Abstract: ***A thyroglossal duct cyst is the most common congenital neck mass which occurs in about 7% of the population due to incomplete obliteration of the thyroglossal duct which normally regresses during fetal development. It typically presents in children but can also persists in adults. These are usually asymptomatic, benign cysts with a good prognosis but can rarely transform to malignancy. About 1% of the thyroglossal duct cysts are malignant and the most prevalent variant is papillary thyroid carcinoma (80%), which has a good prognosis. Here we report a case of 25 year old female who was diagnosed with thyroglossal cyst and underwent Sistrunk's operation and confirmed to have papillary carcinoma of thyroid post operatively. The patient had normal functioning thyroid gland and no lymph node involvement on follow up imaging and was advised regular follow up.***

Keywords: Thyroglossal duct cyst, Papillary carcinoma of thyroid, Sistrunk operation, Hyoid bone

1. Introduction

Thyroglossal duct cysts are the most common congenital neck masses[1,] with an incidence of about 7% in the overall population[2]. The typical presentation occurs in about 70% of children and about 7% in adulthood[3].

A thyroglossal duct cyst is a common congenital anomaly resulting from the incomplete obliteration of the thyroglossal duct, a structure present during embryonic development which normally involutes during fetal development[4]. These cysts typically present as midline neck masses, often in children, but can also be found in adults. While generally benign, thyroglossal duct cysts may occasionally be associated with malignant transformations, which are rare but clinically significant.

The presence of papillary thyroid carcinoma within a thyroglossal duct cyst is uncommon with incidence less than 1% [5], the diagnosis is usually made on incidental evaluation of thyroglossal cyst and confirmed post operatively with histopathology. Papillary thyroid carcinoma accounts for approximately 80% of the malignancy arising from the thyroglossal duct[6]. Papillary thyroid carcinoma (PTC) is the most prevalent and common type of thyroid malignancy, with favorable prognosis compared to other thyroid cancers.

This case report highlights a rare possibility of papillary thyroid carcinoma arising from a thyroglossal duct cyst. The aim is to emphasize the importance of the malignant potential of papillary carcinoma in thyroglossal duct cysts even though it's a benign condition.

2. Case Report

A 25 year old female patient presented with complaints of swelling in the anterior aspect of neck with no history of dysphagia / breathing difficulty / hoarseness of voice / loss of weight and loss of appetite. Patient presented in euthyroid status with no symptoms of hypo or hyperthyroidism and no compressive symptoms. Patient had no significant comorbid conditions and there was no history of radiation or a positive family history. On physical examination there was a swelling in the midline of the neck above the thyroid notch of size 3 x 2 cm, round in shape, smooth surface, margins well defined, soft in consistency, moves with deglutition and protrusion of the tongue. Skin over the swelling normal. No warmth or tenderness. No abnormal pulsations and no dilated veins. No other thyroid nodule or cervical lymph nodes were palpable.

3. Investigations

Thyroid function test revealed euthyroid status. Contrast Enhanced Computed Tomography of Neck done which showed a well defined cystic lesion in midline infrahyoid region in the pre - epiglottic fat. Multiple punctate calcific foci and enhancing solid components within the cystic lesion without any significant cervical lymphadenopathy (Fig 1). The thyroid and parathyroid region appeared normal.

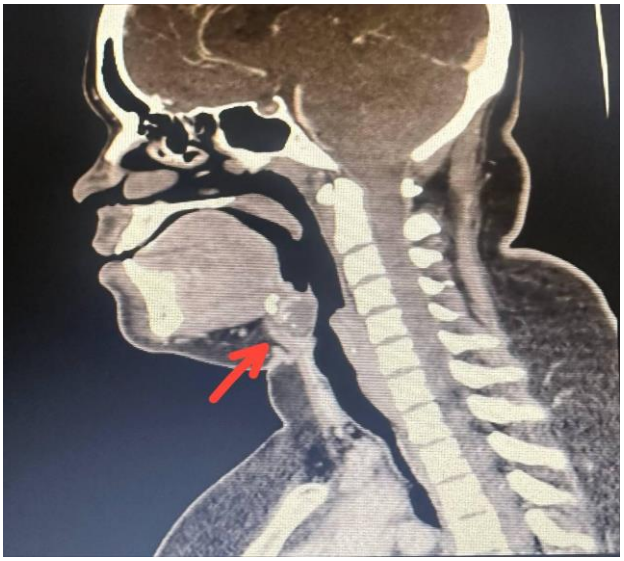

Figure 1: CECT Neck showing a well defined cystic lesion in midline infrahyoid region

Video Laryngoscopy done which showed a localised swelling in the base of epiglottis on left side (Fig 2) and the vocal cords were found to be mobile.

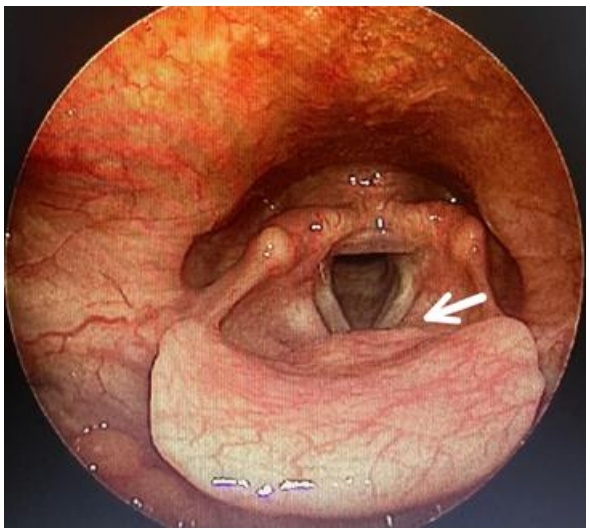

Figure 2: Video laryngoscopy showing bilateral vocal cords and a localised swelling in the base of epiglottis on left side.

A diagnosis of thyroglossal cyst was made based on the history, clinical examination and radiological findings.

Patient underwent Sistrunk's operation under general anaesthesia. Intra operatively after raising the sub platysmal flap and retracting strap muscles, a cyst of size 2 x 3 cm identified just below the hyoid bone extending to the retro hyoid area. Cyst tract behind the hyoid bone traced and removed along with body of hyoid bone (Fig 3) and sent for histopathological examination.

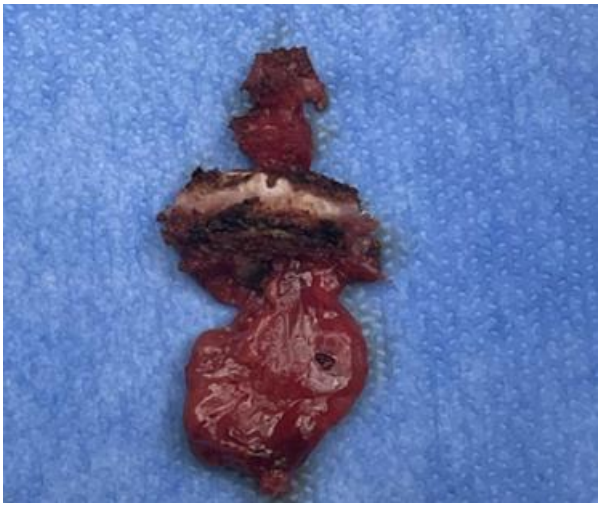

Figure 3: Sistrunk's operation - retro hyoid tract - excised specimen

Histopathological examination showed a unilocular cystic lesion composed of fibrous thickened capsule showing intraluminal complex papillary lesion. The intracystic papillary lesion is composed of varying size arborizing and anastomosing hyalinized fibrovascular cores lined by cuboidal lining epithelial cells with extensive areas of denudation and calcification. The epithelial cells lining the papillae have fibrovascular cores showing nuclear features of papillary thyroid carcinoma associated with stromal hyalinization, fibrosis, degenerative changes and histiocytic reaction. The fibrous wall also showed patchy areas of bony trabeculae with marrow elements and hyaline cartilage and fibromuscular stroma. The fibrous wall showed patchy areas of aberrant thyroid tissue with chronic inflammation. The neoplasm showed focal minimal invasion into the surrounding fibrous capsule but not involving the capsular margin (Fig 4). These findings are suggestive of papillary thyroid carcinoma - classic variant arising from the thyroglossal duct cyst.

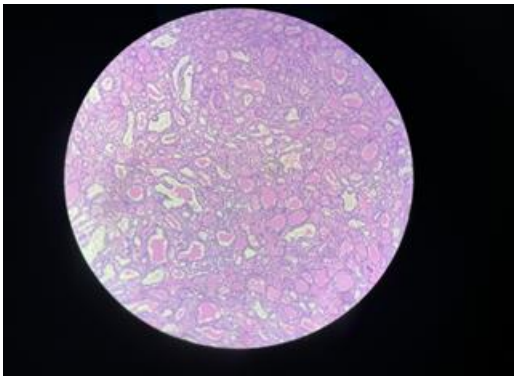

Figure 4: Microscopic picture of papillary carcinoma arising in a thyroglossal cyst.

Repeat ultrasound of the neck was done post operativley which showed normal thyroid functioning in the existing anatomical thyroid gland bed.

4. Discussion

The confirmation of papillary carcinoma of the thyroglossal duct is made by histological examination post operatively. The incidence of papillary carcinoma arising from the

thyroglossal duct cyst is 1% [5] and occuring most commonly in women than men[7]. Thyroglossal duct cyst usually occurs in early adulthood, in patients with malignant potential it occurs in an older age group. The most common type of malignancy arising from the thyroglossal duct cyst is papillary carcinoma (92.1%) followed by squamous cell carcinoma (4.3%). The usual presentation is a midline neck swelling, non tender and soft in consistency. If the cyst is hard, irregular with palpable cervical lymph nodes, it raises a suspicion for malignancy[8]. Even untreated thyroglossal cyst over a long period of time can transform into malignancy. The preferred management of papillary carcinoma arising from a thyroglossal tract cyst is the Sistrunk's procedure and the diagnosis of malignancy is confirmed post operatively by histological examination. Post operatively the thyroid gland is assessed and depending on the extent of the disease, the treatment modalities differ. The management of papillary carcinoma is Total thyroidectomy +/ - neck dissection based on lymph node status. If the cancer cells persist after surgery, radioactive iodine therapy should be considered to reduce the risk of metastasis and to prevent recurrence. Patients are advised life long thyroid hormone supplements.

The prognosis of thyroglossal duct cyst is excellent. About 10% of thyroglossal cyst recur after the Sistrunk's procedure. The recurrence is due to incomplete excision of the thyroglossal tract leaving behind the middle third of the hyoid bone.

Papillary carcinoma of thyroid has a good prognosis compared to other types of thyroid cancers with a five year survival rate of 90% - 95%. Similarly papillary carcinoma arising from the thyroglossal cyst has a good prognosis and it depends on the extent of the disease and appropriate treatment. Early detection and management lead to favourable outcomes. The overall prognosis is good with a survival rate of 99.4% and recurrence rate of 4.3%[9]. Recurrence of papillary carcinoma in a thyroglossal cyst is uncommon but it can metastasise or persist in the thyroid tissue. Hence regular follow up and monitoring with clinical examination and radiological imaging is advised to detect any signs of recurrence and to ensure early detection and management.

5. Conclusion

Thyroglossal cyst is usually a benign condition with malignant transformation being rare, malignancy should be considered when the cyst is hard, fixed or irregular. The presence of papillary carcinoma in thyroglossal cyst is rare but this case suggests the importance of the possibility of malignancy in a thyroglossal cyst. The accurate diagnosis is made with proper history, clinical examination, radiological investigations and confirmed with histopathological examination of the specimen. Early detection with treatment and follow up leads to good outcomes with favourable prognosis in papillary thyroid carcinoma arising from the thyroglossal duct cyst.

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To

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Sub: "UNIFOCAL PAPILLARY CARCINOMA OF THE THYROGLOSSAL TRACT IN A 25 YEAR OLD FEMALE – AN UNCOMMON CLINICAL ENTITY " - reg

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Thank you for submitting the above article write up for a possible publication in an indexed journal. This proposal was reviewed by IEC, on 02/09/2024. The Institutional Ethics Committee approved the write up for journal submission as there was no evidence of plagiarism and the subject in the study were accessed from MRD.

Yours sincerely,

Dr. Ravi Shankar Kannan
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