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RESEARCH ARTICLE

PAPILLARY CARCINOMA OF THYROGLOSSAL DUCT CYST: CASE REPORTS.

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Abstract

Thyroglossal duct cysts are most common etiology of midline neck mass in pediatric population representing around 70%. Although thyroglossal duct cysts are common, cases of thyroglossal duct cyst carcinoma are rare with an incidence of 1%. We presented two cases of female patients who presented to our hospital with asymptomatic neck masses developed gradually over years which were diagnosed with papillary carcinoma after Sistrunk's procedure with total thyroidectomy. Both patients were followed up post-operatively with lab investigations and imaging with no signs of recurrence. Lack of such cases due to its rare entities especially in our region, necessitate more studies to establish clear guidelines for the management of thyroglossal duct cyst carcinoma.

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Introduction:-

Thyroglossal duct cysts (TGDC) are the most common cause of neck mass in childhood, accounting for approximately 70% (1). They can originate anywhere in the neck, but most commonly occur in the midline beneath the hyoid bone, with a reported incidence of 7% (2). Roughly 1% of these develop into a carcinoma, with papillary being the most common histological subtype, representing about 92% of such cases (3, 4). One study reviewed all cases since 1915 and deduced that TGDC carcinomas have a prevalence of around 1% (5). They mostly present asymptotically with a history of a neck mass present for many years, usually since childhood (6). However, a small number of cases have presented with pain and dysphagia. Here we report on two cases of TGDC carcinomas because of its extremely rare occurrence, and the minimal reporting in our country.

Case Reports:-

Case 1

A 45-year-old female patient had thyroglossal cyst for more than 10 years which was increasing in size recently with no dysphagia, shortness of breath, or hoarseness of voice. The patient complains of otalgia intermittently.

On examination, a 2x2 hard mass was felt in the neck, which was firm, non-mobile and fixed to underlying tissue, with smooth borders and no associated skin changes. The mass moved with protrusion of the tongue. Thyroid function test was within normal limits.

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Ultrasound (US) was done and showed a heterogeneous well-defined oval right thyroid nodule measuring 0.8x0.8 cm, and a well-defined echogenic oval non-shadowing left lobe nodule measuring 0.5x0.4 cm [figure 1]. It also revealed a left paramedian cervical lesion measuring 1.9 x 1.4 x 2 cm, which was cystic with an irregular solid component and increased vascularity. No cervical lymphadenopathy was noted.

A Computed Tomography (CT) scan of the neck showed a well-defined heterogeneous lesion measuring 1.8x1.5x1.7 cm with mixed soft tissue and fluid densities seen in the left para-midline location just anterior to the thyroid gland with no clear invasion [figure 2]. It also showed multiple hypodense nodules in the thyroid with the largest seen in the right lobe. Fine needle aspiration (FNA) raised the suspicion of papillary thyroid carcinoma [figure 3]. It showed a thyroglossal cyst with thyroid tissue inside, that had features of a papillary thyroid carcinoma.

A Sistrunk's procedure and total thyroidectomy were done in 2014. The pathology report confirmed the diagnosis of papillary cell carcinoma in the thyroglossal duct cyst and multi-nodular goiter. The tumor measured 1.1 cm, infiltrating the surrounding fibroadipose tissue and muscle with negative margins. The patient received 100 mci of Iodine 131 ablation postoperatively and subsequent scans were normal, including a whole-body scan. After that, the patient was put on levothyroxine 125 mcg. Follow up of the patient was achieved through stimulated thyroglobulin (TG) and TG antibody. After 3 years of monitoring, the patient has not shown any signs of recurrence.

Case 2

A 20-year-old female patient was complaining of midline neck mass for 7 months. The patient has no history of dysphagia or shortness of breath. A negative family history or radiation therapy. On examination, a submental hard mass measuring 2x2 cm was felt with an otherwise unremarkable examination. The mass moved with swallowing and tongue movement.

The US showed a midline, heterogenous, well-defined thyroglossal cyst, with internal calcification and no obvious vascularity, measuring 3.0 x 3.4 x 3.5 cm [figure 4]. Also, the thyroid gland showed multiple bilateral hypoechoic nodules with mild vascularity, and multiple prominent lymph nodes in the neck were noticed.

The CT scan of the neck revealed a lesion with coarse calcification and faint central enhancement in the infrahyoid area with continuation to the thyroid gland, suggesting complicated thyroid cyst with probable malignant degeneration [figure 5]. Also, multiple enlarged lymph nodes were noted at level-1 and level-2 with the largest one at level-2 A. Two years later, it showed an increase in the size of the cyst with more lobulation [figure 6]. FNA was done and revealed focal features indicative of papillary carcinoma [figure 7]. The right lobe showed papillae lined by atypical cells with crowded nuclei, optical clearing, nuclear grooving and numerous psammoma bodies.

A total thyroidectomy with excision of the thyroglossal cyst was done in 2016. The pathology report confirmed the diagnosis of papillary thyroid carcinoma with tumor size of 1.4 cm in the right lobe with no extra thyroid extension or lymphovascular invasion. In addition, papillary thyroid carcinoma of thyroglossal duct cyst was diagnosed as a result of the tumors extension.

Discussion:-

Normally, the thyroid gland starts to develop from the base of the neck. Eventually, it migrates to its normal location below the thyroid cartilage leaving behind a track connecting them, which is usually obliterated by the 5th to 10th week of gestation. Failure of closure leads to the formation of TGDC, and the factors responsible for this are still unknown. However, the remnant can also be a duct, fistula, or ectopic tissue within a cyst or duct (10).

TGDC adenocarcinomas are rare entities, with an incidence of around 1%. There are two main histopathological subtypes, which are papillary and squamous cell carcinomas. Papillary carcinomas account for 94 % of these cases, while less than 5% represent tumors of squamous origin (7). The proposed mechanisms for occurrence is either de novo synthesis or secondary from an occult thyroid origin (8). The majority of cases seem to support the de novo theory, which is identified on histopathology by the presence of thyroid tissue devoid of parafollicular cells that would result in a medullary carcinoma. Some believe that the duct can act as a canal for the metastasis of thyroid carcinomas (9).

Since TGDC carcinomas usually present as painless neck masses, which are usually found incidentally, clinical diagnosis is difficult. Therefore, a pathological analysis is essential in confirming the diagnosis. A thorough clinical history and physical examination, in conjunction with radiological studies help to define the mass' characteristics, the extension to the adjacent structures, and distant metastasis. A mural mass with calcification and positive enhancement on imaging would raise the suspicion of malignancy (11). Preoperative fine needle aspiration has a controversial role in diagnosis with a sensitivity of 62% and positive predictive value of 69% (12).

Management of such cases is dependent on the histopathological examination. Some reports recommend a more conservative approach, which is mainly by performing the Sistrunk procedure. A total thyroidectomy is recommended for patients over 45 years of age, with advanced lesions with extracapsular invasion or spread to the lymph nodes, concerning radiological features, and prior radiation exposure (13). However, others have recommended a more invasive method of treatment, which entails performing total thyroidectomies, in addition to the Sistrunk procedure. This was due to their postoperative findings, which revealed concomitant thyroid carcinomas in most their cases 63.2 % (14). A radioactive iodine ablation should be considered in cases of concurrent thyroid and TGDC cancer, large tumors, or lymph node involvement (13).

In our cases, the preliminary diagnosis was achieved via FNA and an invasive approach was applied as a management, even in the younger patient. Post-operative pathology confirmed the diagnosis of papillary carcinoma. In comparison, a report from Riyadh of a similar case of papillary carcinoma (15). They followed the same management and performed bilateral neck dissection, which ruled out other malignant foci. They also gave their patient adjuvant radio iodine ablation and thyroid suppression therapy. After regular follow up, their patient showed no signs of recurrence at 5 years.

So, it is important to remain vigilant in patients with TGDC, even though carcinomas may be rare. Also, clear guidelines for the management of such cases should be formed to minimize the risk of unnecessary total thyroidectomies and undiagnosed cancers.

Conclusion:-

In summary, TGDC is quite common with only 1% developing into cancer. Clear guidelines for the management of TGDC carcinomas need to be established to improve patient care.

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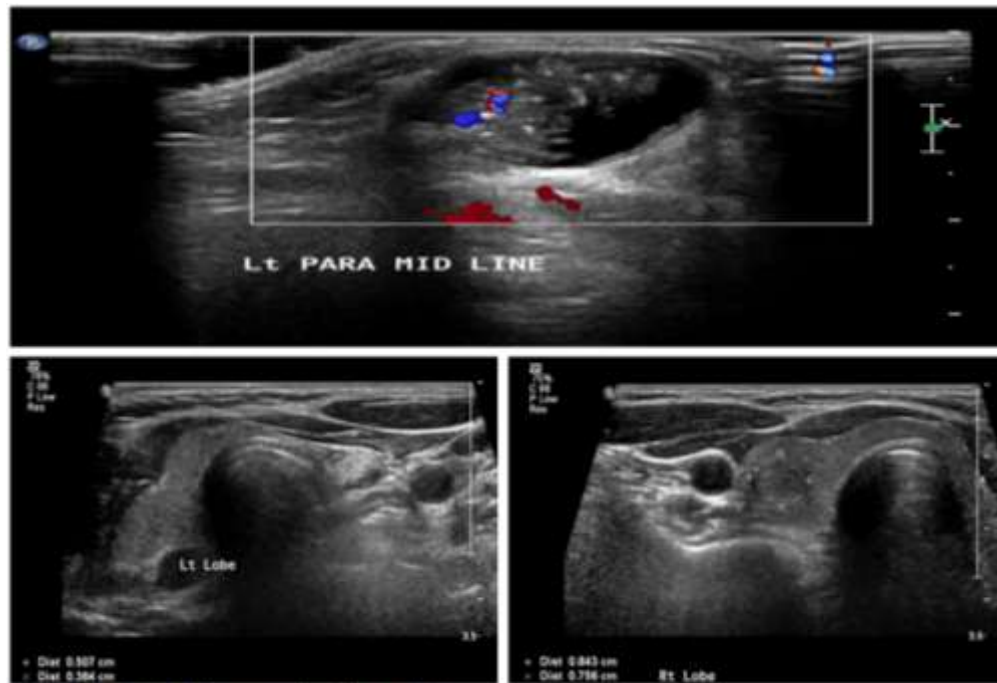


Figure 1: Neck Ultrasound of the nodule

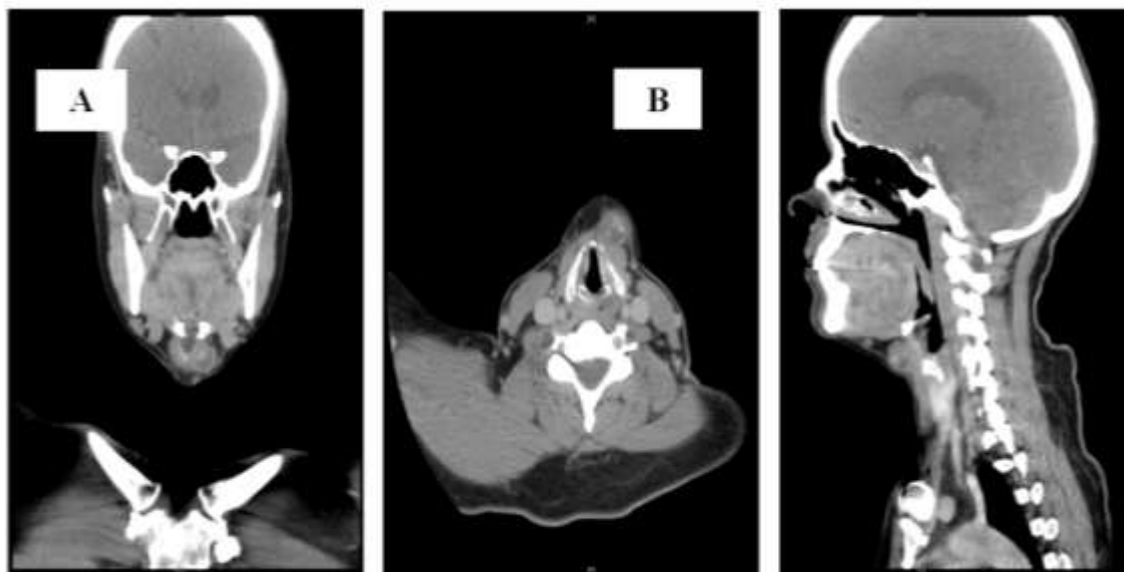


Figure 2: Neck CT of lesion

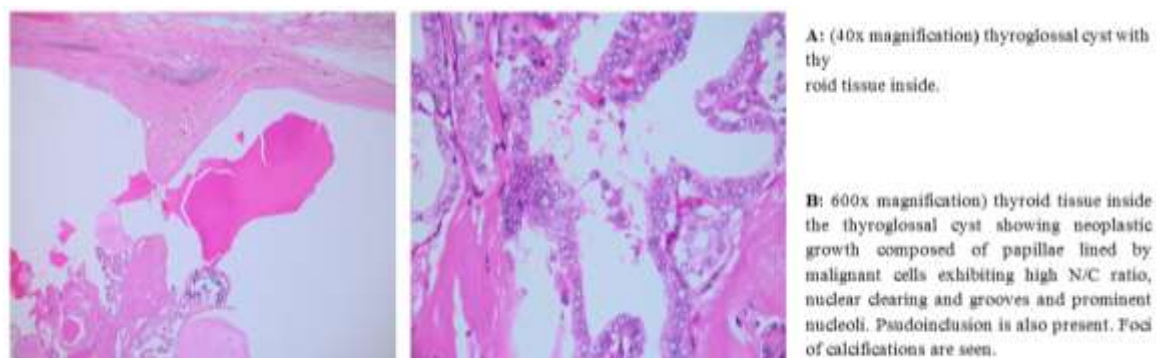


Figure 3: FNA of the lesion

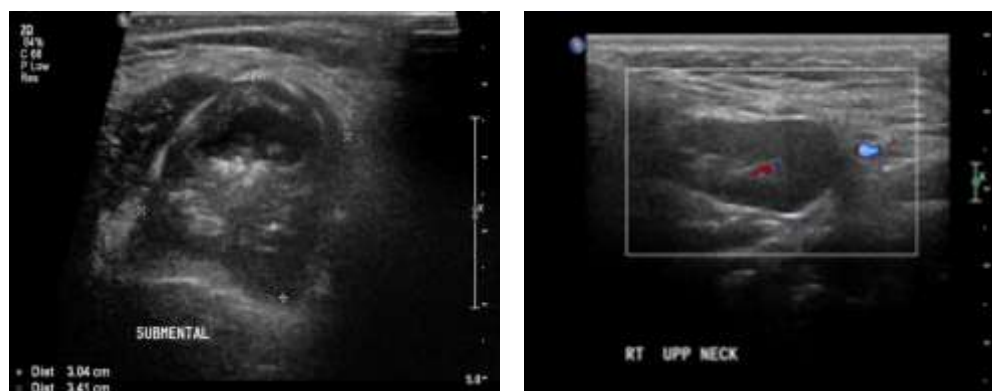


Figure 1: Neck US of the nodule

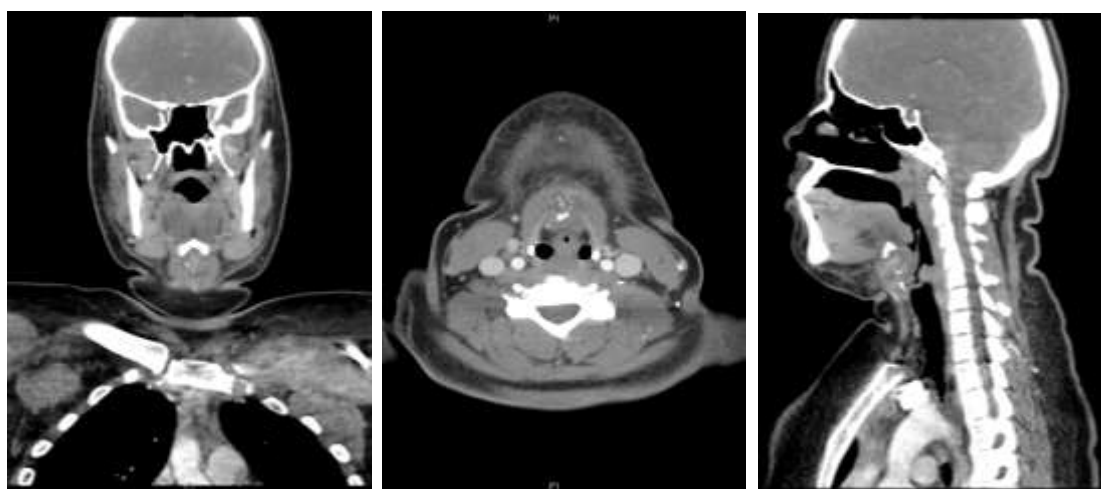


Figure 2: Neck CT scan of the lesion

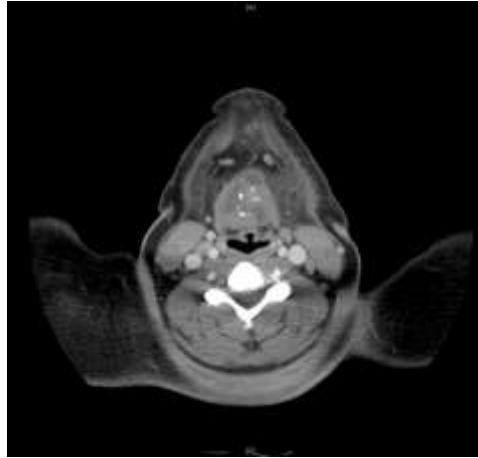
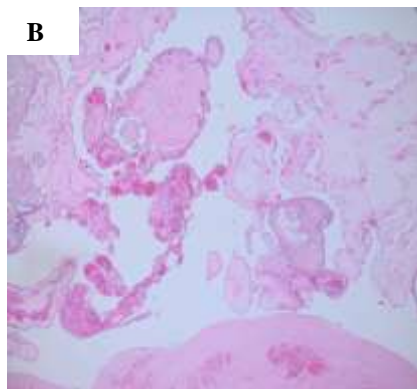
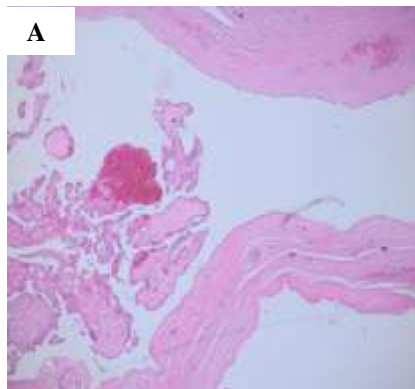


Figure 3: Neck CT showing progression two years later



A: (40x) papillary thyroid carcinoma involving cyst with fibrous wall and reactive lymphoid tissue.

B: (40x) foci of calcification "one feature of papillary thyroid carcinoma".

C: (600x) papillary thyroid carcinoma

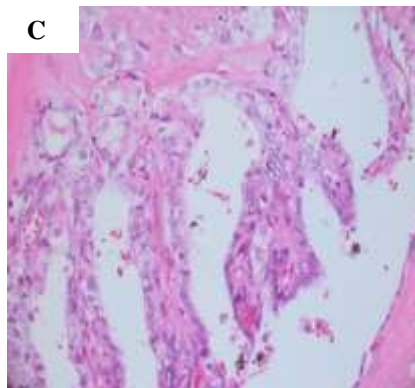


Figure 4: FNA of thyroid nodule