

## PRIMARY PAPILLARY CARCINOMA OF THYROID IN PERSISTENT THYROGLOSSAL DUCT CYST IN ADULT

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### ABSTRACT

Thyroglossal duct cyst is the most common congenital anomaly of the neck. Occurrence of papillary carcinoma in the thyroglossal duct cyst in the presence of normal thyroid gland is rare. A 50 year old female presented with an anterior midline neck swelling since childhood. CT scan neck showed a multiloculated cystic mass extending from isthmus region of thyroid to hyoid bone with a centrally placed soft tissue lesion with scattered calcification. Thyroid gland appeared normal in size and parenchymal attenuation in its usual location. The mass was excised. Cut section showed a multiloculated cystic lesion having a small solid nodule with papillary excrescence. Microscopic examination showed features of papillary carcinoma of thyroid. Patient underwent total thyroidectomy with post operative radioactive iodine treatment. Histopathological examination of the thyroidectomy specimen revealed no evidence of malignancy. No evidence of recurrence during 18 months follow up.

**KEYWORDS:** Primary Papillary Carcinoma of Thyroid, Thyroglossal Duct Cyst

### INTRODUCTION

Thyroglossal duct cyst (TGDC) is the most common anomaly in thyroid development. TGDC occurs in the midline neck and is diagnosed clinically<sup>(1)</sup>. About 1% of the thyroglossal duct cyst are histologically malignant and 80% of these are papillary carcinoma of thyroid gland type<sup>(2)</sup>. Brentano was first to describe this condition in 1911, and since then about 215 cases have been reported in the literature<sup>(5)</sup>. TGDC carcinoma may be clinically indistinguishable from benign TGDC, and the diagnosis in most cases is incidental after surgical resection. The use of fine needle aspiration cytology under ultrasound guidance may enhance the preoperative diagnosis<sup>(4)</sup>. The prognosis of papillary carcinoma arising in a thyroglossal cyst is similar to that of papillary carcinoma of the thyroid gland having cure rates in excess of 95%<sup>(5)</sup>. Here we report a case of primary papillary carcinoma of thyroid arising in the thyroglossal duct cyst in a 50 year old female.

### CASE REPORT

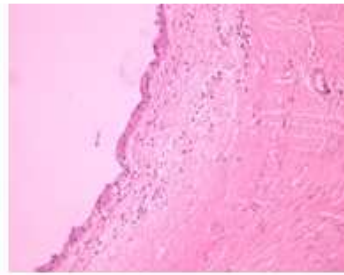
A 50 year old lady presented with a slowly growing midline upper neck swelling since childhood. FNAC was done two years back, but patient lost the report. Thyroid function tests were normal. CT scan of neck showed a multiloculated cystic mass measuring 55x31x42mm extending from isthmus region of thyroid till hyoid bone. A centrally placed soft tissue lesion with scattered calcification seen within it. Thyroid gland was in its usual location with normal size and parenchymal attenuation. The mass was excised and sent for histopathological study.

Grossly the mass measured 5x5x3cm. Cut section revealed a multiloculated cyst containing thin jelly like fluid. One small solid grayish white nodule of size 5mm was seen in one septa with surface excrescences (figure 1).

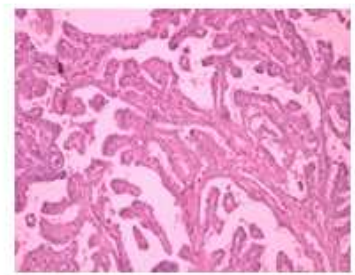
Microscopic examination of the cyst wall showed the cystic spaces lined by flattened to squamous epithelial cell (figure 2). Sections from septal nodule showed features of papillary carcinoma of thyroid. Tumor cells are arranged in complex arborising papillary structure with nuclear overcrowding, nuclear clearing, grooving and intranuclear pseudo-inclusion. Foci of calcification seen (figures 3,4,5,6). Following the histopathological report total thyroidectomy was done with postoperative radioactive iodine therapy. Detailed histopathological examination of the excised thyroid gland revealed no evidence of malignancy (figure 7). Patient is doing well without any clinical evidence of recurrence or metastasis after 18 months of follow up.



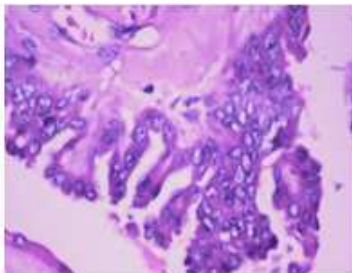
**Figure 1: Multiloculated Cyst with the Small Nodule(Arrow)**



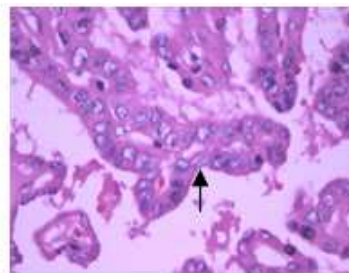
**Figure 2: (H&E,100x) Cyst Wall Lined by Squamous Epithelium**



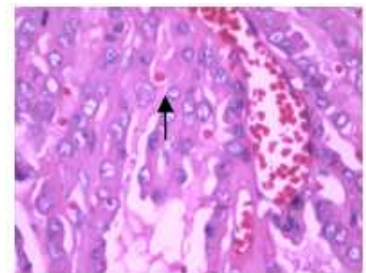
**Figure 3: H&E,100x) Complex Papillary Structures**



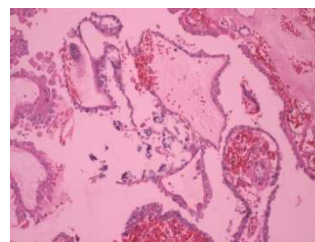
**Figure 4: (H&E,400x) Nuclear Overcrowding and Clearing**



**Figure 5: (H&E,400x) Nuclear Grooving (Arrow)**



**Figure 6: (H&E,400x) Intranuclear pseudo-inclusion(arrow)**



**Figure 7: (H&E,100x) Calcification**

## DISCUSSIONS

The clinical diagnosis of thyroglossal duct cyst is usually accurate due to its anatomical location. Complications like infection or sinus formation are common. But malignant transformation is rather rare<sup>(3)</sup>. More than 200 cases of malignant tumors in TGDC have been reported. There are two types of thyroglossal duct carcinomas: thyrogenic carcinoma and squamous cell carcinoma. The results in the literature show that papillary carcinoma is the most common type (80%), followed by “mixed” papillary-follicular carcinoma(8%) and squamous cell carcinoma(6%). The remaining

6% include rare cases of Hurthle cell, follicular and anaplastic carcinomas<sup>(2)</sup>.

In patients with thyrogenic carcinomas, the possibility of metastasis from occult primary thyroid tumor exists. Windstrom et al suggested two histopathologic diagnostic criteria for primary carcinoma of the thyroglossal cyst: the localization of the carcinoma to a clearly demonstrable thyroglossal duct cyst and the absence of carcinoma on histopathologic examination of the thyroid gland<sup>(2)</sup>. In our case also the subsequent total thyroidectomy specimen did not reveal any evidence of malignancy, thus fulfilling the diagnostic criteria proposed by Windstrom et al.

The most common clinical presentation is the presence of an anterior neck mass indistinguishable from that of a benign TGDC. However, malignancy must be suspected in cases of TGDC in light of the recent changes in the clinical features like large or increasing size, hard, fixity, irregular shape, previous exposure to ionizing radiation<sup>(1,4)</sup>. Imaging diagnostic techniques (ultrasonography, scintigraphy and CT scan) are usually unable to preoperatively diagnose a malignant disease and fine needle aspiration cytology yields a correct result in only 66% of the cases<sup>(4)</sup>.

There is a great deal of controversy regarding the surgical treatment of TGDC carcinoma. Although some surgeons consider the Sistrunk procedure alone to be sufficient for patients with no signs of disseminated diseases<sup>(6)</sup>, recent reports recommend a total thyroidectomy in addition to the Sistrunk procedure. The number of reported incidences of primary thyroid carcinoma, concomitant with TGDC carcinoma is between 11 and 33%. The foci of such carcinomas are small (ranging from 0.2 cm to 1.5 cm), and are frequently not palpable or detectable by preoperative imaging techniques. A total or subtotal thyroidectomy has been recommended if there is cystic wall invasion by the carcinoma, or if the TGDC carcinoma is greater than 1.0cm, because larger lesions are more likely to behave aggressively<sup>(4)</sup>.

Weiss and Orlich also found papillary carcinoma foci in the thyroid gland in 11.4% cases of TGD cyst carcinoma. They recommended total thyroidectomy following the histopathological diagnosis of papillary carcinoma in the TGD cyst, despite the thyroid gland was normal in the initial assessment. Patel et al. did not recommend additional treatments in low-risk patients. Patients below 45 years old of age, with tumor diameter <4 cm, and no sign of extensive metastasis were low risk<sup>(7)</sup>. Our patient was 50 years old and total thyroidectomy was done in spite of a normal clinical and radiological thyroid gland.

The prognosis for papillary TGDC carcinoma is excellent, with occurrence of metastatic lesions in less than 2% of cases. Follow-up procedures consist of physical examinations, ultrasound of the surgical region and thyroid, and a total body scintigraphy<sup>(4)</sup>.

## CONCLUSIONS

Formation of thyroid carcinoma in a thyroglossal cyst is a rare entity, with controversial management options. Preoperative diagnosis is often difficult. Hence extensive grossing is essential to avoid an underdiagnosis. Its behavior and prognosis are usually similar to that of the papillary thyroid cancer. Our case was no exception and the carcinoma was not discovered until the histological examination of the excised cyst.

There are very limited cases of the primary TGD cyst carcinomas and more limited follow up reports in the literature. We should consider a possible malignancy in the TGD cyst although it is very rare. When we diagnose a TGD remnant we should strongly recommend the patient a Sistrunk procedure because the possibility of the malignancy increases by age. We also should evaluate the thyroid gland for a possible malignancy.

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