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Case Report

Pathology Section

Latent Primary Papillary Microtumor in Thyroglossal Duct Cyst Wall - A Rare Case Report with Review of Literature

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ABSTRACT

Thyroglossal duct cyst (TDC) is a common congenital midline neck mass. Generally TDCs are benign and incidence of malignancy in TDC is less than 1%. Majority of the tumors arise from thyroid remnants in the wall of the cyst and the common histologic type is papillary carcinoma.We report a 25-years-old man diagnosed as TDC based on clinical and radiological findings. Histopathological examination confirmed TDC with an incidental intracystic latent papillary microtumor. The papillary microtumor was confirmed with Immuno histochemical study using Cytokeratin-19 (CK 19) marker. The present case report emphasizes the histopathological features and prognostic significance of papillary microtumor in TDC with review of literature.

Keywords: Thyroglossal duct cyst, Papillary micro tumor, Cytokeratin

INTRODUCTION

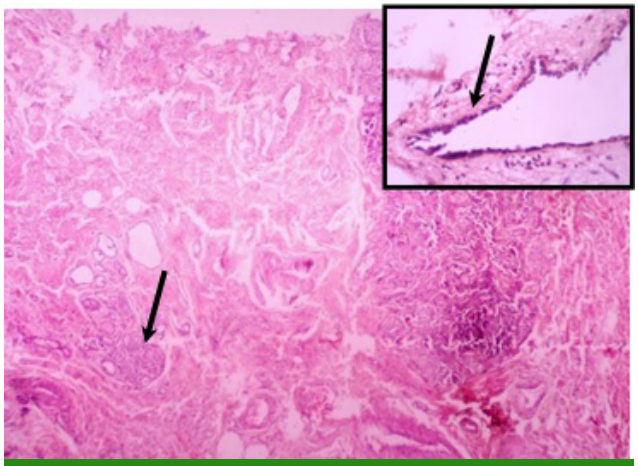
TDC is a developmental cyst arising from incompletely atrophied thyroglossal duct. Being a benign entity it is extremely rare to show a malignant tumor [1]. However if malignancy occurs, it arises from the cysts epithelium or from thyroid remnants in its wall [2]. Tumors reported in TDC are: Classical papillary carcinoma (81.7%), Mixed papillary & follicular carcinoma (6.9%), Squamous cell carcinoma (5.2%), Follicular and Adenocarcinoma (1.7% each), and Anaplastic carcinoma (0.9%) [3]. There are no previous documented reports of latent papillary microtumor in TDC. Hence, this could be probably the first case report of this rare entity. The histopathological characteristics with prognostic significance of the microtumor is also been discussed.

CASE REPORT

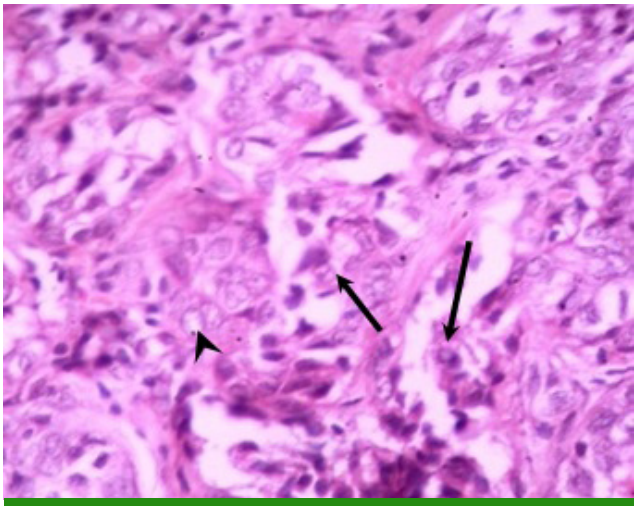
A 25-years-old man presented with a 5x4 cms cystic swelling in front of the neck extending below the hyoid bone. The swelling moved with deglutition. There was no visible thyroid swelling clinically and no lymphadenopathy. Routine lab tests reports were within normal limits. With a clinical diagnosis of Thyroglossal Duct Cyst, patient was subjected to Sistrunk's operation. The excised cyst was sent for Histopathological examination. Microscopic examination showed cyst wall lined by various cells comprising of pseudo stratified columnar, cuboidal and squamous epithelial cells at places with underlying inflamed stroma [Table/Fig-1]. Focal areas within the cyst wall showed follicular cells arranged in microfollicles [Table/Fig-1], solid pattern and tiny nests with nuclear clearing, grooving and pseudo-inclusions [Table/Fig-2]. Histopathological diagnosis of latent papillary microtumor in a TDC was made and confirmed with cytoplasmic positive Cytokeratin-19 marker [Table/Fig-3].

DISCUSSION

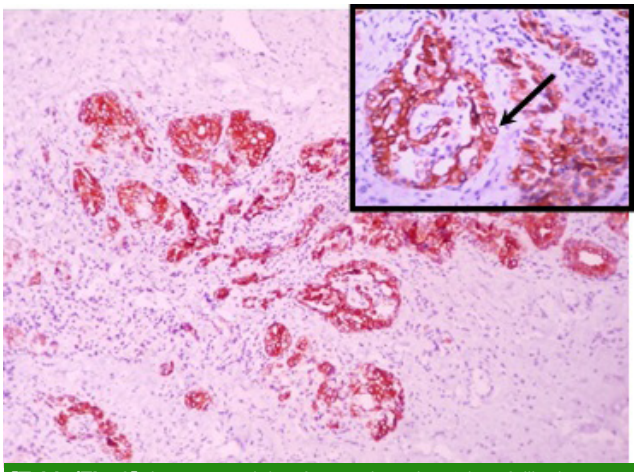
TDC carcinoma is rare and is seen in less than 1% of cases [4]. Less than 250 cases have been reported from different centers since 1911 [5]. Carcinoma arising from TDC has two origins: Thyrogenic carcinoma from thyro embryonic remnants in the duct or cyst and squamous cell carcinoma arising from metaplastic squamous cells that line the duct.



[Table/Fig-1]: Photomicrograph showing Thyroglossal cyst wall with a focus of neoplastic follicles in cluster [Arrow]. (H&E x 50). Inset shows cyst wall lined by cuboidal cells [Arrow, Inset]. (H&E, x 400)



[Table/Fig-2]: Photomicrograph showing follicular tumor cells with characteristic oval optically clear nucleus [Arrow-Head]. There are nuclear crowding, grooves [Short-Arrow] and occasional Pseudo inclusion [Long-Arrow]. (H&E x 400)



[Table/Fig-3]: Immunostaining for cytokeratin 19 in a follicular lesion of Thyroglossal cyst wall shows diffuse cytoplasmic reaction in tumor cells. (Cytokeratin x 100). Inset [Arrow] shows diffuse intense cytoplasmic reaction around optically clear nucleus. (Cytokeratin x 400)

Most of the TDC malignancy arises from thyroid remnants and the common histologic type is papillary carcinoma. However twin malignancies of papillary carcinoma and squamous cell carcinoma in TDC have been reported. Classical papillary carcinoma usually presents in the third to fifth decades of life with a female preponderance, while latent papillary microtumor lacks the female predominance. The risk factors for thyroid papillary carcinoma are irradiation and pre existing thyroid diseases, but papillary microtumor in TDC arises denova [1]. Gross morphology of typical papillary carcinoma is whitish firm granular lesion with ill defined margins. In our case, gross morphology revealed cyst wall with inner smooth surface. There was no obvious thickening of the cyst wall and no solid whitish firm mass lesion. So far reported papillary carcinoma in TDC have shown classical microscopic morphology of branching papillae with fibro vascular core and nuclear features of papillary malignancy in the form of nuclear clearing, grooving, crowding and pseudo inclusions. In our case, Microscopic examination showed solid, microfollicular and tiny nests of follicular epithelial cells with nuclear crowding, clearing, grooves and nuclear pseudo inclusions. The above classical morphology favors the histopathological diagnosis of incidental latent papillary microtumor of TDC. The gold standard for the diagnosis of follicular thyroid lesions, particularly papillary carcinoma, is Histological examination. However in order to confirm the diagnosis in a tumor, which did not show the classic papillary pattern, Immunohistochemistry was done. The sensitivity and specificity using Cytokeratin-19 as a single marker is reported as high as 92%, & 97% respectively [6] and hence, CK-19 marker test was done. Normal follicles, benign papillae of graves' disease and nodular hyperplasia are negative for CK-19 [12]. Though benign follicular lesions like follicular adenoma & carcinoma, reactive follicular cells in thyroiditis shows positive expression of CK-19, the distribution is focal and less intense [6]. In our case all neoplastic follicular cells showed strong & diffuse immuno reactivity for CK-19 which confirmed the histopathological diagnosis. Prognosis of papillary carcinoma arising in the TDC is good and is similar to that of thyroid papillary carcinoma having cure rates in excess of 95%. But cases of invasive papillary carcinoma of TDC and with nodal metastasis have been reported [3]. In latent papillary microtumor most of the cases remain dormant and do not grow to clinically apparent disease. Thus, the prognosis is excellent. Treatment of papillary carcinoma in TDC depends upon tumor size, local invasion to adjacent structures, thyroid and nodal metastasis. Sistrunks operation followed by suppressive dose of thyroxin and regular follow up is recommended for incidental papillary carcinoma in TDC without invasion [6]. In latent papillary microtumor regular follow up is sufficient if it is incidentally diagnosed after Sistrunks procedure (avoid over treatment).

This case is reported in view of its rarity, non aggressive behavior and to highlight the distinct morphological features. Regular follow up alone is sufficient for this asymptomatic incidental tumor type.

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