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Case Report

Pathology Section

Primary Papillary Carcinoma Arising in Thyroglossal Duct Cyst: A Rare Case Report

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ABSTRACT

The most common anomaly in thyroid development is thyroglossal duct cyst which results from the failure of involution of the thyroglossal tract. Carcinoma arising from the thyroglossal duct is rare with fewer than 275 cases reported till date. Papillary carcinoma is the most common among them. Majority of patients are females within first two decades of life. Five year survival is less than 2%. We report a case in 27 year-old female who presented with painless, midline neck swelling which slowly progressed in size over one year. Histopathology revealed primary papillary carcinoma arising from the thyroglossal cyst.

Keywords: Papillary carcinoma, Thyroglossal duct cyst, Thyroid

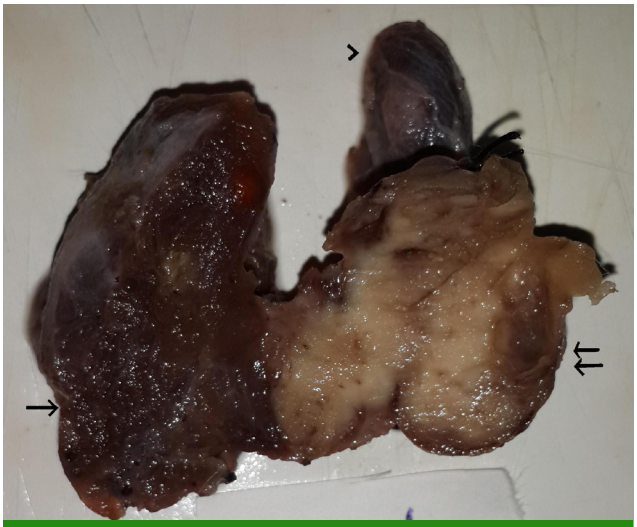
INTRODUCTION

Thyroglossal duct cyst is the most common anomaly in thyroid development and usually presents within the first two decades of life. The usual presentation is as an enlarging painless midline neck mass [1]. Carcinoma arising from the thyroglossal cyst is very rare with fewer than 275 cases reported till date and usually cannot be distinguished clinically from benign cysts. The final diagnosis is established following histopathological examination [1,2].

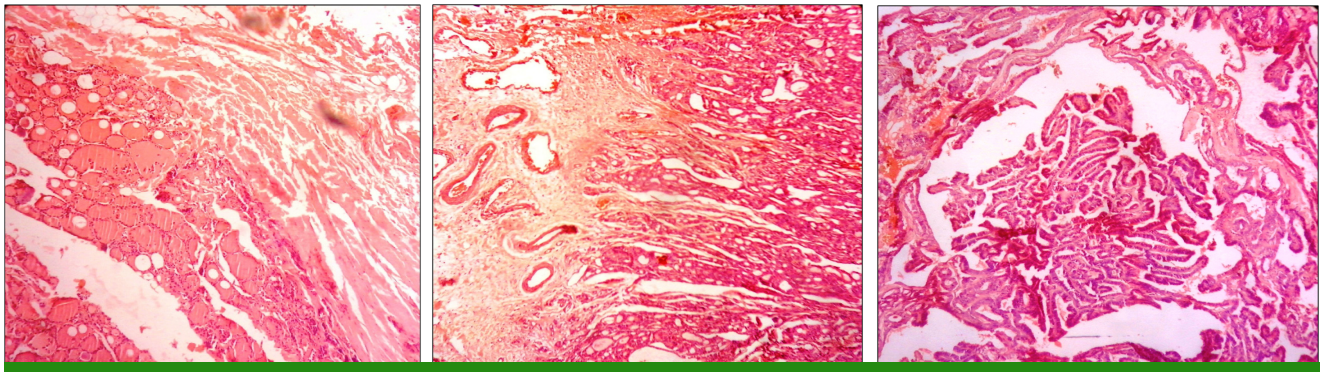
CASE REPORT

A 27-year-old female presented with painless, midline neck swelling since one year which slowly progressed in size. No history of change in voice or difficulty in swallowing noted. On clinical examination, the swelling measured 4x3 cm and moved with deglutition and protrusion of tongue with no evidence of cervical lymphadenopathy. A clinical diagnosis of multinodular goiter with thyroglossal duct cyst was given. Thyroid scan revealed features of nodular goitre of right lobe of thyroid with a mid-line swelling at the level of thyroid cartilage which did not show functioning thyroid tissue, possible radiological diagnosis of multinodular goitre with thyroglossal duct cyst was given. Fine Needle Aspiration of the nodule showed thyroid follicular cells having features of isonucleosis, scanty cytoplasm arranged in follicular pattern and in discrete in a background of abundant colloid and plenty of cystic macrophages. A cytological diagnosis of nodular goitre with cystic change was offered. Hematological/biochemical investigations and thyroid function tests were within normal limits. Sistrunk procedure with right hemithyroidectomy was performed.

Gross examination of the specimen showed right lobe of thyroid tissue along with thyroglossal tract and a grey white solid to cystic mass at the isthmic end of the thyroglossal tract. Thyroid tissue measured 4.5x2x1 cm and cut section was gray brown. Gray white mass attached to thyroglossal tract measured 4x3x1 cm and cut section was gray white [Table/Fig-1]. Histopathological examination of right lobe of thyroid showed features of nodular goitre. Thyroglossal tract showed fibrocollagenous tissue without lining epithelium and subepithelial stroma showed normal thyroid tissue [Table/Fig-2]. Sections from gray white mass with adjacent thyroglossal



[Table/Fig-1]: Gross photograph showing right lobe of thyroid (single arrow), thyroglossal tract (Arrow head) and carcinoma in thyroglossal duct cyst (double arrow)



[Table/Fig-2]: Microphotograph showing fibrocollagenous wall of thyroglossal tract with stroma showing normal thyroid tissue. H&E 100X
[Table/Fig-3]: Microphotograph showing tumour arising from fibrocollagenous wall of thyroglossal duct cyst having papillary and follicular pattern. H&E 100X
[Table/Fig-4]: Microphotograph showing papillary structures with features of papillary carcinoma. H&E 100X. (Images from left to right)

tract shows fibrocollagenous wall lined by flat epithelial cells and tumour shows features of papillary carcinoma having both papillary and follicular architecture [Table/Fig-3,4]. Final histopathological diagnosis of primary papillary carcinoma of thyroglossal duct cyst was made. Patient was followed up and total thyroidectomy was performed after one month. The specimen did not show tumour tissue. No sign of recurrence was noted on follow up of patient for eight months.

DISCUSSION

Thyroglossal duct cyst is the common anomaly in the development of thyroid. It is due to failure of the duct to involute completely and lead to the development of a thyroglossal duct cyst due to the accumulation of secretions which is seen in about 7% of adult population [2,3]. Carcinoma developing in the thyroglossal duct cyst are very rare with an incidence of 0.7 to 1%. It was first described by Brentano in 1911 and Uchermann in 1915. The etiology is attributed to radiation exposure, metastasis from an occult primary thyroid neoplasm or may be de novo from the ectopic thyroid tissue. It is common in women than men (3:2) and in second decade of life (2^{nd} to 6^{th} decade reported) [3,4]. In the present case it was female in third decade.

The usual symptom is a midline neck swelling which moves with deglutition and protrusion of tongue for six months to six years. Recent increase in size, irregular shape, hard consistency, hoarseness of voice and dysphagia usually raise the suspicion of a neoplastic transformation. FNAC diagnosis will be a cystic lesion or can yield positive results in 66% of the cases [3,5]. In the present case painless swelling in the midline of neck which moved with deglutition and protrusion of tongue was the clinical presentation. FNAC diagnosis was nodular goitre.

Histopathology gives confirmative diagnosis. The criteria for the diagnosis of primary carcinoma of the thyroglossal duct include demonstration of the duct or cyst wall with epithelial lining, subepithelial stroma showing normal thyroid follicles, tumour tissue in lumen of duct/cyst and no sign of primary carcinoma in the thyroid gland. The most common thyroglossal duct carcinoma is papillary carcinoma (75-80%), but other tumour types such as mixed papillary-follicular carcinomas (7%), squamous cell carcinoma (5%), follicular carcinoma (1.7%), Hurthle cell carcinoma and anaplastic carcinoma (0.9%) have been reported. Prognosis of a well differentiated carcinoma in thyroglossal cyst is excellent while that of squamous cell carcinoma or anaplastic carcinoma is poor [6-8]. In the present case histopathology revealed thyroglossal duct with no lining epithelium but with subepithelial thyroid tissue. The thyroglossal cyst structure along with features of papillary carcinoma of papillary/follicular pattern was very well evident.

Sistrunk procedure for the removal of the thyroglossal duct and cyst is the treatment of choice with a cure rate of 95% in a radiologically normal thyroid. However, Sistrunk procedure with total thyroidectomy has been suggested by many authors if the tumour size is larger than 1 cm, cyst wall or thyroid is involved and age of the patient is more than 40 years. It should be followed by radioiodine ablation and thyroid stimulating hormone suppression therapy. Cervical lymph node metastasis calls for a radical or modified radical neck dissection [4,8,9]. In the present case the patient underwent right hemithyroidectomy with Sistrunk procedure as the pre-operative diagnosis was nodular goitre with thyroglossal duct cyst. However after the histopathological diagnosis total thyroidectomy was performed.

About 4% of thyroglossal duct carcinomas are found to be locally invasive and 11% show lymph node metastasis [1]. Papillary carcinoma of the thyroglossal cyst has risk of metastasis in less than 2% of cases, however majority of the cases on follow-up did not show recurrence [1,2,8,9]. Papillary carcinoma is known for its multicentric/multifocal growth and lymphatic spread and about 10% of the patients with thyroglossal cyst carcinoma have been found to have microscopic foci of carcinoma in thyroid gland [10]. Thyroidectomy is therefore, recommended similar to papillary carcinoma thyroid because of their common embryological origin [11,12]. Thyroidectomy

also aids in staging, detection of metastases and recurrence. Follow up is recommended [2,3,4]. In the present case, the thyroid tissue did not show tumour deposits and there was no cervical lymphadenopathy. Patient was followed up for eight months which was uneventful.

CONCLUSION

Although papillary carcinoma of thyroglossal duct cyst is rare, one should be aware of the possibility of malignancy in a thyroglossal duct cyst. Though it has good prognosis, a long-term follow-up is mandatory because of recurrence and metastasis.

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