

## Case Report

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# Primary papillary carcinoma in thyroglossal cyst

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### Abstract:

*Introduction: Thyroglossal cyst is the most common congenital midline neck swelling. Most of the cases are benign with rare malignant transformation is approximately in 1-2% cases. Only about 200 cases have been reported from different centers since 1911. Clinical presentation is similar to that of benign cyst and diagnosis is usually made incidentally during histopathological examination of excised cyst.*

*Case report: We report a case of 46-year-old woman who underwent Sistrunk's operation for thyroglossal cyst. Histopathological examination revealed papillary carcinoma. Thyroxine was added in suppressive dose and patient was on regular follow-up for 2 years. No thyroid abnormality was found preoperative cytology, scintigraphy as well as per-operative procedure and post-operative follow-up period.*

*Conclusion: Sistrunk's operation followed by suppressive dose of thyroxin and regular follow up is recommended for thyroglossal duct cyst carcinoma in asymptomatic incidental case without the cyst wall invasion.*

**Key words:** Thyroglossal cyst, papillary carcinoma, Sistrunk's operation.

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### Introduction:

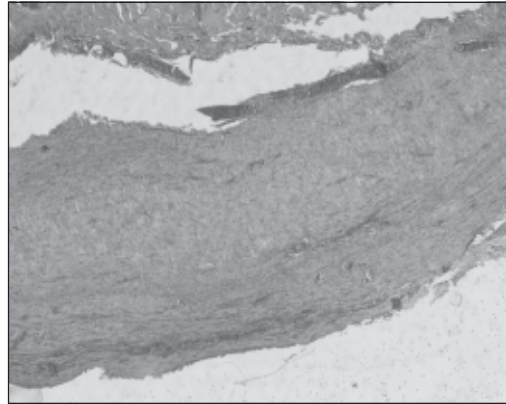
Thyroglossal cyst is the most common congenital midline neck swelling arising from dilatation of the embryonic remnant epithelium during thyroid migration. Most of the cases are benign with rare malignant transformation in approximately 1-2% cases.<sup>1, 2, 3</sup> Benign thyroglossal cyst is common in pediatric age group but reported malignant cases are usually in adult. Approximately 200 cases have been reported sporadically from different centre since 1911 and papillary carcinoma is the commonest histo-pathological variety.<sup>4</sup> Indistinguishable symptoms between benign and malignant conditions, preoperative diagnosis is difficult but carcinoma may be suspected when the cyst is rapidly enlarged hard, irregular presented in the adult group. Incidentally diagnosis is usually made post operatively during histopathological examination.<sup>5</sup>

To the available literature review no thyroglossal cyst carcinoma yet reported from Bangladesh. Here we report a case of 46-year-old woman with primary papillary carcinoma in thyroglossal cyst and discuss the diagnosis and management of this rare case.

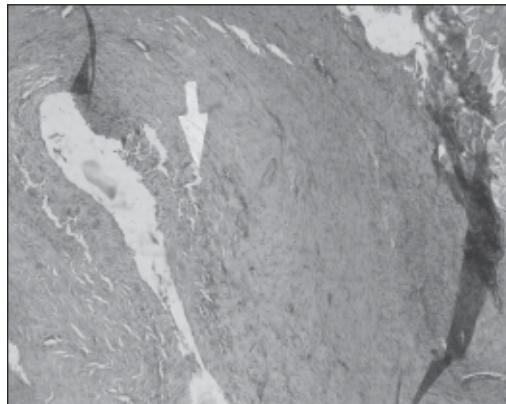
#### Case report:

A 46-year-old woman presented with complaint of painless swelling in front of the neck for 12 years. The swelling was gradually increasing in size but for the last 6 months, it was enlarging rapidly for which she attended the consultant otolaryngologist. She had no history of difficulty in swallowing except slight discomfort feeling. She had no respiratory distress or change of voice. Physical examination revealed a cystic swelling in the thyrohyoid region which moves on swallowing as well as protrusion of tongue. The swelling was globular in shape, firm in consistency, well defined margin, measured about 4X3 cm, smooth surface, non-tender. There was no palpable neck node. FNAC revealed hemorrhagic cystic colloid goiter. Ultrasonogram of the thyroid gland noticed extra thyroid cystic swelling with normal thyroid gland. CT scan revealed a large hypodense space occupying lesion with patchy calcification in the thyroid region reported as cystic goiter. So, investigation raises diagnostic dilemma whether it was cystic colloid goiter or thyroglossal cyst. But when it was diagnosed clinically as thyroglossal cyst, Sistrunk's operation was done. Per-operatively a thick walled cyst was found between two lobes of thyroid gland which was attached with the hyoid bone and isthmus by a cordlike structure.

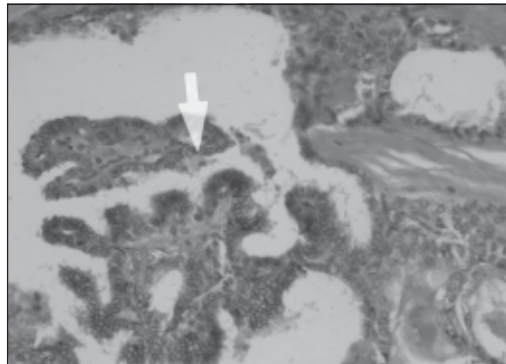
Excised cyst was sent for histopathological examination where the diagnosis was confirmed as papillary carcinoma of thyroglossal cyst.



**Figure 1:** Showing cyst wall (H&S stainX10)



**Figure 2:** Showing cyst wall and tumour (H&S stainX10)



**Figure 3:** Showing focus of papillary projection (H&S stainX10)

Suppressive dose of the thyroxine was added after 7 days and advised to follow up 3 monthly for 1<sup>st</sup> year, 6 monthly for 2<sup>nd</sup> year

and yearly in next consecutive years. She was followed up for 1<sup>st</sup> 2 years as per advice. During follow up besides clinical examination USG of the thyroid gland, serum TSH & thyroglobulin level was estimated and no abnormality was detected and she had been passing healthy life. But she was lost for follow up after 2 years.

#### Discussion:

A thyroglossal cyst carcinoma was first reported in European literature in 1910 by Ucherman and it finally made into English literature in 1927 by Dr Owen and Colleagues. Though thyroglossal cyst wall contains thyroid tissue in 60% of the cases reported incidence of concomitant primary thyroid carcinoma and thyroglossal duct cyst carcinoma is between 11.0-33.0%.<sup>6</sup> Clinical presentation of thyroglossal duct cyst carcinoma is similar to benign condition but recent and rapid enlargement raises suspicion of malignant transformation.

In this case the patient had asymptomatic swelling in front of the neck for twelve years with recent six months history of rapid enlargement and discomfort in the neck. No palpable neck node was found.

FNAC has low sensitivity for the detection of thyroglossal cyst malignancy.<sup>1</sup> Imaging tests (Ultrasound and CT scan) are not conclusive for preoperative diagnosis. In this case FNAC and CT scan reports were misleading as both reports suggested as cystic colloid goiter.

Most of the reported cases are confirmed incidentally in histopathological examination of the excised cyst<sup>5, 7, 8, 9, 10, 11</sup> and of them 75-85% of cases were established as papillary adenocarcinoma.<sup>5</sup> Though clinically benign, postoperative histopathological examination of this case revealed papillary carcinoma.

Controversies exist in relation to rational and effective therapeutic approach for thyroglossal

cyst carcinoma. After Sistrunk's operation some prefers total thyroidectomy<sup>1, 2, 3, 14</sup> followed by suppressive dose of thyroxine but some opine total thyroidectomy have no significant impact on the outcome.<sup>5, 7, 11 - 13</sup> But if any nodule is detected in the thyroid gland by palpation or ultrasonogram and cyst wall is found invaded by carcinoma then total thyroidectomy followed by I<sup>131</sup> ablation and suppressive dose of thyroxine therapy is advocated after Sistrunk's operation.

In this case we avoided total thyroidectomy & radioiodine ablation. After Sistrunk's procedure we advised suppressive dose of thyroxine to avoid thyroidectomy and radioiodine related morbidity.

#### Conclusion:

Though there is no definite treatment protocol for thyroglossal duct cyst carcinoma, in asymptomatic incidental case without the cyst wall invasion Sistrunk's operation followed by suppressive dose of thyroxin and regular follow up is recommended.

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