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## PAPILLARY CARCINOMA IN THYROGLOSSAL CYST: CASE SERIES

### ABSTRACT

Thyroglossal cyst is one of the common problem in ENT practice. Sometime the cyst can harbor malignancy which also presents like a normal thyroglossal cyst. It is only after the post-operative histopathological examination we can diagnose it. Management of thyroglossal cyst carcinomas has always been a controversial for the need of total thyroidectomy and adjuvant treatment. Here we present two different cases of thyroglossal cyst carcinoma which were managed differently.

**Keywords:** Papillary carcinoma, Thyroglossal cyst, Thyroidectomy

### INTRODUCTION

Thyroglossal cyst (TG) is one of the common problem identified in ENT practice, however the papillary carcinoma in cyst is relatively uncommon entity. Since it is rare disease there is no fixed guidelines to deal with such kind of problem. The first case of thyroglossal duct cyst carcinoma was described by Brentano in 1911.<sup>1</sup> Total number of reported cases in world literature till date is 275.<sup>2</sup> Beside the papillary carcinoma, follicular carcinoma and squamous cell carcinoma has also been reported in the TG cyst. The differentiated thyroid cancer in TG cyst behaves aggressively than regularly presenting papillary carcinoma of thyroid. Since there are no fixed guidelines to treat papillary carcinoma in TG cyst, there are lot of controversies regarding its management especially the need of total thyroidectomy and radioactive ablation. After routine Sistrunk operation for TG cyst, post-operative histopathological surprises the surgeon as cyst being reported as the papillary carcinoma. Surgeon have to decide the need for total thyroidectomy with or without radioactive ablation standing on some scientific evidence which we are going to discuss in later part of the discussion. Here we present two patients of papillary carcinoma reported in the TG cyst and the difference in management of these patients.

### Case 1:

35 years male farmer presented with midline neck swelling for 3 years, insidious and gradually progressive without any pressure symptoms. It was 3x2cm<sup>2</sup>, soft in consistency moving with deglutination and protrusion of tongue at the level of hyoid bone with normal overlying skin.

Ultrasonography of neck shows lobulated complex cystic 2.9x2.2x2.2 cm<sup>3</sup> lesion with internal debris without calcification but with intralesional vascularity. It was lying below hyoid going in between hyoid and thyroid cartilage into pre-epiglottic space. The thyroid gland was located in normal position. Fine needle aspiration cytology was consistent with thyroglossal cyst.

With this classical picture of thyroglossal cyst he underwent Sistrunk operation, however his post-operative histopathology was reported as papillary carcinoma with lymphovascular invasion. The size of lesion was 3.5x1.5 cm<sup>2</sup> and hyoid bone was free of disease. Neck ultrasonography was repeated after two weeks of surgery which did not reveal any lesion in the thyroid gland and surgical site was normal. Since there was extensive lymphatic invasion in post-operative histopathology, total thyroidectomy was done. Second post-operative histopathology

report did not show any focus inside the thyroid. He was discharged with supplement thyroxine, which was titrated according to TSH value.

### Case 2:

A 59 years old male, retired officer presented in ENT out patient department with history of painless swelling in the anterior neck for last 15 years. The swelling was constant and had progressed gradually over a duration of 15 years. The swelling was moving with deglutination and protrusion of tongue. There was no skin changes or other constitutional symptoms associated with the swelling. Ultrasonography (USG) revealed approximately 3x3 cm<sup>2</sup> complex cystic space occupying lesion in the subhyoid region suggestive of thyroglossal cyst with normal thyroid gland. Fine needle aspiration cytology showed follicular variant of papillary carcinoma in the cyst. He underwent wide local excision (WLE) with removal of body of hyoid along with isthmusectomy. Post-operative histopathological report showed papillary carcinoma in thyroglossal cyst of size 6mm without any lymphovascular invasion (Figure 1). He was advised for follow up every 6 monthly.

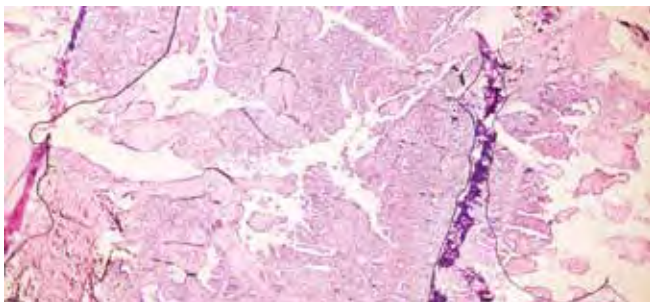


Fig.1: Microscopic Picture of the Papillary Carcinoma in Thyroglossal Cyst

### DISCUSSION:

The incidence of papillary carcinoma in TG cyst is around 1.6 %.<sup>3,4</sup> Histologically, papillary thyroid carcinoma is by far the most common, followed by mixed papillary and follicular carcinoma and squamous cell carcinoma. Medullary thyroid carcinoma has never been reported. The pathogenesis of papillary carcinoma in TG cyst is also debatable.

Metastasis of occult papillary carcinoma from thyroid to the cyst is one of theory<sup>5</sup>, hence total thyroidectomy once the papillary carcinoma is reported in TG cyst is justified. We did the same for our first case where papillary carcinoma was reported in histopathological report but surprisingly no focus was identified in final

report later on. Data base from Memorial Sloan-Kettering Cancer Center shows that thyroidectomy was performed after the diagnosis of thyroglossal duct cyst carcinoma in 30 out of the 62 patients. Out of 30 thyroidectomy specimen, 22 (73.3%) showed either benign or normal tissue and papillary carcinoma was seen in 8 (26.7%).<sup>6</sup> So the risk of total thyroidectomy over the benefits in cases of TG cyst carcinoma has to be justified. Therefore it has to be done by the expert who can minimize the morbidity of total thyroidectomy like recurrent laryngeal nerve palsy and hypocalcemia.

Another theory of pathogenesis of papillary carcinoma in TG cyst states that carcinomas in the thyroglossal duct/cyst arise de novo in the thyroid remnants present in the thyroglossal cyst.<sup>7</sup> This explains why medullary carcinoma thyroid has never been reported in thyroglossal tract remnants as medullary carcinoma arises from parafollicular cells. Hence thyroid specimen after total thyroidectomy for papillary carcinoma in the TG cyst may not yield malignant lesion as in our first case. Papillary carcinoma in TG cyst present as a regular presenting TG cyst in the midline neck. These cyst on routine USG and FNAC may not show any suspicious features of malignancy. FNAC has sensitivity of 50–60% in diagnosing carcinomas of thyroglossal duct cyst as compared to primary thyroid gland lesions where the sensitivity is 85%.<sup>8</sup> Management of TG cyst papillary carcinoma is still controversial as we don't have fixed guidelines and recommendation. The main controversy is for total thyroidectomy. Literature is divided, however, broad guidelines indicate that total thyroidectomy should be done based on risk stratification. High-risk factors include age >45 years, male, size of TG cyst papillary carcinoma >4cm, extra-capsular invasion of excised cyst, positive nodal status, history of irradiation, presence of cold nodules in thyroid on thyroid scan.<sup>9</sup> Detection of focus in thyroid in preoperative USG neck may help to decide for thyroidectomy. However, there is good chance of missing the lesion as they are very tiny according to Bakkar et al.<sup>10</sup> He reported 43% risk of missing a malignancy in sonographically normal thyroids after Sistrunk's surgery in cases of papillary carcinomas in TG cysts.

Low-risk cases are; age <45 years, cyst size less than 4 cm, no extra capsular infiltration and no neck node. Sistrunk's operation only will suffice but requires regular and close follow up.

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