

# Papillary Thyroid Carcinoma Arising in Thyroglossal Duct Cyst and Locally Infiltrated to Hyoid Bone: Issue of Individualized Management

<sup>1</sup>Shahnaj Hossain Dinu, <sup>2</sup>Fatima Begum, <sup>3</sup>Shamsun Nahar Bailey, <sup>4</sup>Jasmin Ferdous and <sup>5</sup>Zeenat Jabin

<sup>1</sup>MD (Nuclear Medicine) Resident, BSMMU

<sup>2</sup>Professor & Head, Thyroid Division, National Institute of Nuclear Medicine & Allied Sciences (NINMAS)

<sup>3</sup>Assistant Professor & SMO, NINMAS

<sup>4</sup>Associate Professor & PMO, NINMAS

<sup>5</sup>Director, Institute of Nuclear Medicine & Allied Sciences (INMAS), ShSMCH

**Correspondence Address:** Dr. Shahnaj Hossain Dinu, NINMAS, Block-D, BSMMU campus, Shahbag, Dhaka-1000

Email: drshahnajhossaindinu@gmail.com

## ABSTRACT

Thyroglossal duct cyst (TGDC) is the most common developmental anomaly of the thyroid gland, while TGDC carcinoma with local infiltration to the hyoid bone is extremely rare. The reported case of papillary thyroid carcinoma (PTC) was diagnosed in TGDC along with local infiltration to the hyoid bone and needed individualized management. A 70-year-old male patient presented with a history of PTC arising in TGDC and locally infiltrated to the hyoid bone followed by total thyroidectomy and radical sistrunk operation and was referred to National Institute of Nuclear Medicine and Allied Sciences (NINMAS) for radioactive iodine therapy (RAIT). His post-operative ultrasound of the neck revealed an irregular shaped, fungating, grossly non-homogenous mixed echogenic mass lesion invading the hyoid bone, measuring about 25mm X 28 mm. Radioactive iodine (<sup>131</sup>I) of 150 mCi was administered because the residual hyoid bone mass was inoperable and he was on thyroxine suppression therapy. The post-therapy <sup>131</sup>I scan (RxWBS) showed two intense foci of radiotracer concentration (RTC) in the upper area (above the thyroid bed) and an intense focal RTC in the fundal region of the stomach. The activity in the stomach region was evaluated by ultrasound imaging, which revealed a soft tissue mass or thickening of the mid-region of the posterior wall of the stomach (3.3 cm X 1.6 cm). A CT scan of the chest was also performed to rule out metastasis, and it revealed multiple nodular lesions in both lung fields (most likely secondary) as well as tiny subcentric lymphnodes in the lower pre- and right paratracheal regions after nearly one month of RAIT when the patient was stable for examination. EBRT is still under consideration as adjuvant therapy. Tyrosine kinase inhibitors (TKI) may be considered in cases of refractoriness to radioiodine and disease progression.

**Keywords:** Thyroglossal duct cyst, Papillary thyroid carcinoma, Sistrunk operation, Total thyroidectomy, Radioactive iodine therapy (RAIT), EBRT, Tyrosine kinase inhibitor (TKI).

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## INTRODUCTION

Thyroglossal duct cysts (TGDC) are the outcome of the persistence and cystic growth of the remnants of the thyroglossal tract.

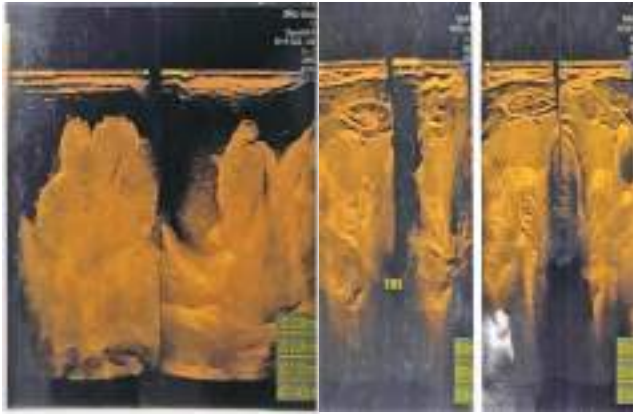
TGDC is a common anomaly that affects 7% of the population (1). The rate of carcinoma in the TGDC is approximately 1%, but infiltration of the hyoid bone is extremely rare (2). Its management is also controversial. The objective of reporting this case is to highlight the rarity of papillary thyroid carcinoma (PTC) in a TGDC with an almost unresectable hyoid bone metastasis and the dilemma of its individualized management.

## CASE REPORT

A 70-year-old male patient presented with an anterior midline neck mass which was soft, non-tender, and situated in the midline below the hyoid bone, with a normal thyroid gland and no enlarged cervical lymph nodes on palpation. High resolution neck ultrasound revealed two hyperechoic heterogeneous, cauliflower-like solid masses on the hyoid bone, measuring approximately 3 cm X 2.2 cm and 1.5 cm X 1.4 cm, respectively (Figure 1.a), with no mass in the thyroid gland exhibiting normal echotexture (Figure 1.b).

A contrast enhanced CT scan of the neck revealed carcinoma valliculae (5 cm X 3 cm X 3 cm) with destruction of the hyoid bone, invasion of the left-sided base of the tongue, and extension anteriorly in the submental space (Figure 2).

Ultrasound guided fine-needle aspiration cytology (FNAC) from the midline swelling was done, and that reported malignant salivary gland neoplasm / metastatic thyroid carcinoma. Total thyroidectomy and radical sistrunk operation were done by the head and neck surgeons and the histopathology report revealed papillary thyroid carcinoma in TGDC.

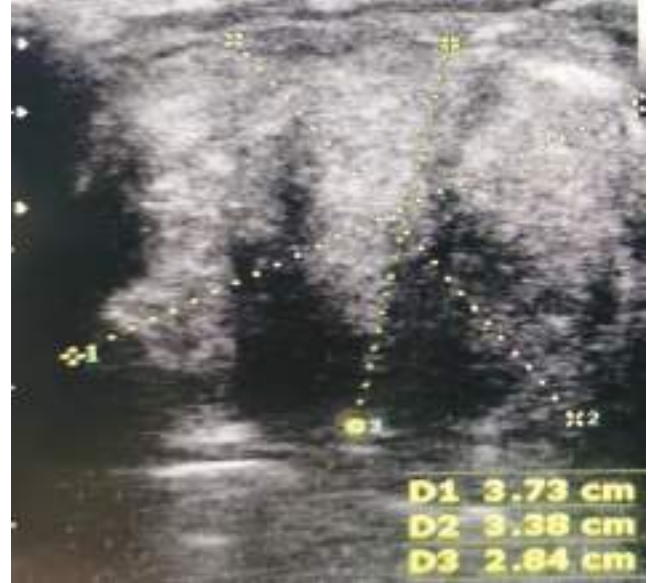


**Figure 1. a) High resolution ultrasound image of neck showing two hyperechoic cauliflower like solid masses on the hyoid bone measuring about 3 cm X 2.2 cm and 1.5 cm X 1.4 cm. b) Normal sized thyroid lobes with uniform tissue echopattern are visible in the same patient.**



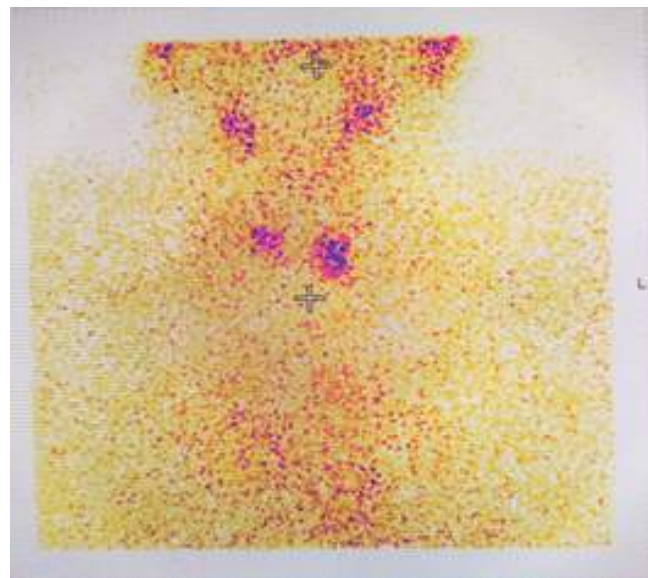
**Figure 2: Axial view of CT at hyoid bone level showing mass invading the hyoid bone.**

However, the resected thyroid gland was unremarkable. This patient was referred to NINMAS for radioactive iodine therapy (RAIT). Post-operative high-resolution ultrasound (HRUS) of the neck revealed no thyroid tissue in the thyroid bed. However, an irregular shaped, cauliflower like, grossly non-homogeneous, mixed echogenic mass lesion was found invading the hyoid bone, measuring about 37 mm X 34 mm X 28mm (Figure 3).



**Figure 3: High resolution ultrasound image of neck. A fairly big, irregular shaped, heterogeneous mass occupying the hyoid bone area and appears to be invading the bone.**

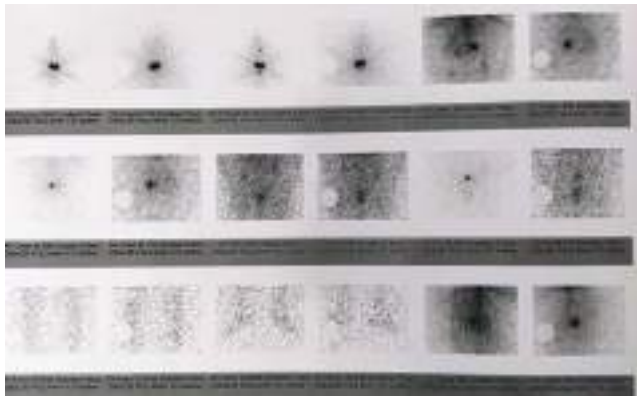
Pre-therapy laboratory investigations revealed serum TSH = 69.99 mIU/L, serum thyroglobulin (Tg) >300 ng/ml, anti Tg antibody (TgAb) < 1.30 U/ml. Technetium (<sup>99m</sup>Tc) thyroid scan showed focal areas of radiotracer concentrations (RTC) in the thyroid bed (Figure 4).



**Figure 4: Thyroid scan showing focal areas of radiotracer concentration.**

A radioactive iodine (<sup>131</sup>I) dose of 150 mCi was administered as further surgery for residual hyoid bone mass was not possible.

The post-therapy  $^{131}\text{I}$  scan (RxWBS) revealed two intense foci of RTC in the upper part (above the thyroid bed) and one intense focal RTC in the fundal region of the stomach (Figure 5).



**Figure 5: Post therapy whole body scan showing focal intense radiotracer concentration above the thyroid bed corresponding to the residual mass and fundal region of stomach.**

This activity persisted even after drinking plenty of water and a repeat scan the next day. On the 7th posttherapy day, the patient complained of blood vomiting. A CT scan of the chest was also performed to rule out metastasis, and it revealed multiple nodular lesions in both lung fields (most likely secondary) as well as tiny subcentric lymph nodes in the lower pre- and right paratracheal regions after nearly one month of RAIT when the patient is stable for examination (Figure 6).



**Figure 6: CT image of both lung fields (axial view) showing multiple nodular lesions.**

The activity in the stomach region was evaluated by ultrasound imaging, which revealed a soft tissue mass or

thickening in the mid-region of the posterior wall of the stomach (3.3 cm X 1.6 cm) (Figure 7).



**Figure 7: Ultrasound image of abdomen. An irregular shaped, solid, heterogeneous mass like lesion is seen at the mid-region of posterior wall of stomach.**

## DISCUSSION

The thyroglossal duct connects the thyroid gland to the foramen cecum via an epithelial junction. During the eighth to tenth week of gestation, the duct is usually obliterated. If the duct fails to completely involute, the residual epithelium can result in the formation of a TGDC. These cysts are the most common developmental anomalies, usually located in the midline, anterior region of the neck, with no apparent clinical symptoms. Ellis and van Nostrand suggested that this failure to involute occurs in approximately 7% of the population (1,3). Complications of these swellings are rare, among these, thyroglossal duct cyst carcinoma (TGDC) is extremely rare, accounting for approximately 1% of all cases, and its management is greatly debated, particularly when the thyroid gland is clinically normal (2, 4). TGDC has an unknown cause. The dominant theories are either metastatic illness from an occult source or spontaneous proliferation from an ectopic origin (5).

Patients with TGDC should have a thorough preoperative evaluation, which includes a thorough physical examination, an accurate head and neck examination, palpation of the thyroid gland, thyroid function tests, a thyroid scan, and a CT or MRI scan. If the cyst becomes hard, fixed, irregular, or suddenly expands with palpable neck lymph nodes, malignancy should be suspected. Meanwhile, a CT or MRI scan may reveal a solid mass with invasive characteristics such as local invasion, hyoid



bone infiltration, and cyst calcification (6, 7). The use of FNAC as a preoperative diagnostic for malignancies is now common. Only a few cases of cancer in TGDC were detected preoperatively (7). Preoperatively, the reported case had a morphologically normal thyroid gland in the HRUS, a normal CT scan, and normal biochemical tests. Postoperative histopathology detected no abnormalities of the thyroid gland either.

The definitive management of TGDC carcinoma is still debatable, with concerns about the therapeutic plan for the thyroid gland and neck dissection. The most common surgical procedure for thyroglossal duct cyst carcinoma is the Sistrunk operation, which involves removing the entire cyst and the central portion of the hyoid bone. A core of tissue around the thyroglossal tract opens into the oral cavity at the foramen cecum (8, 9). The standard Sistrunk's procedure is sufficient for patients with non-metastatic disease who are medically low-risk, but high-risk patients who have intrathyroidal metastases, local invasion, or nodal metastases should undergo an additional total thyroidectomy with neck dissection, as well as postoperative radioactive iodine ablation therapy and suppressive thyroxine therapy (9). The prognosis for papillary TGDC carcinoma is favorable, with metastatic lesions occurring in less than 2% of cases (10). In this case, the patient underwent a total thyroidectomy, Sistrunk surgery, and a radical neck dissection followed by radioactive iodine therapy due to evidence of hyoid bone involvement. The reported case of PTC arising in the TGDC and locally infiltrating the hyoid bone is rare. After the Sistrunk operation, significant tissue was found in the region of the hyoid bone, as evaluated by HRUS of the neck. RAIT was the next option in the presence of the huge, inoperable mass. Suspected metastasis was found post-RAIT and evaluated by RxWBS as well as by a CT scan of the chest and ultrasound imaging of the whole abdomen. The patient was advised to consult a gastroenterologist and a clinical oncologist. The patient

is on suppressive thyroxine therapy, and regular follow-up will be carried out in the thyroid division of NINMAS. EBRT is still under consideration as adjuvant therapy. Tyrosine kinase inhibitors (TKI) may be considered in cases of refractoriness to radioiodine and progression of disease.

## REFERENCES

1. Pacheco-Ojedal L, Micheau C, Stafford N, Marandas P, Luboinski B, Martinez AL. Papillary carcinoma in thyroglossal duct remnants. *European archives of oto-rhino-laryngology*. 1991 Jul;248:268-70.
2. Yang YJ, Hagher S, Wanamaker JR, Powers CN. Diagnosis of papillary carcinoma in a thyroglossal duct cyst by fine-needle aspiration biopsy. *Archives of pathology & laboratory medicine*. 2000 Jan;124(1):139-42.
3. Ellis PD, Van Nostrand AP. The applied anatomy of thyroglossal tract remnants. *The Laryngoscope*. 1977 May;87(5):765-70.
4. Iftikhar H, Ikram M, Nathani KR, Muhammad AY. Papillary thyroid carcinoma within thyroglossal duct cyst: case series and literature review. *International Archives of Otorhinolaryngology*. 2018 Jul;22:253-5.
5. Pietruszewska W, Wągrowska-Danilewicz M, Józefowicz-Korczyńska M. Papillary carcinoma in thyroglossal duct cyst with uninvolved thyroid. Case report and review of the literature. *Archives of Medical Science*. 2014 Oct 23;10 (5):1061-5.
6. Glastonbury CM, Davidson HC, Haller JR, Harnsberger HR. The CT and MR imaging features of carcinoma arising in thyroglossal duct remnants. *American journal of neuroradiology*. 2000 Apr 1;21(4):770-4.
7. Papavramidis TS, Zisi A, Tzorakoleftheraki SE, Koletsa T, Pliakos I, Panidis S, Kotsovolis G, Manani C, Kita M, Michalopoulos A. Papillary carcinoma arising from the pyramidal lobe of the thyroid gland—Two case reports. *Journal of Clinical and Translational Endocrinology: Case Reports*. 2018 Mar 1;7:1-4.
8. Patel NN, Hartley BE, Howard DJ. Management of thyroglossal tract disease after failed Sistrunk's procedure. *The Journal of Laryngology & Otology*. 2003 Sep;117(9):710-2.
9. Kwon JK, Lee SM, Lee HM, Lee JC. Papillary thyroid carcinoma arising from a primary thyroglossal duct cyst with cervical node metastases. *Thyroid*. 2012 Mar 1;22(3):330-1.
10. Park MH, Yoon JH, Jegal YJ, Lee JS. Papillary thyroglossal duct cyst carcinoma with synchronous occult papillary thyroid microcarcinoma. *Yonsei Medical Journal*. 2010 Jul 1;51(4):609-11.