

## Case Report

# Thyroglossal duct papillary carcinoma presenting as submental neck node

Babu Manohar, Raees Abdurahiman\*

Department of ENT and Head and Neck Surgery, Apollo Hospitals, Chennai, Tamil Nadu, India

**Received:** 17 June 2019

**Revised:** 26 July 2019

**Accepted:** 30 July 2019

**\*Correspondence:**

Dr. Raees Abdurahiman,

E-mail: raees023@gmail.com

**Copyright:** © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

### ABSTRACT

Neck node in sub mental area is not uncommon. It occurs mostly due to infective or malignant pathologies in oral cavity. Submental neck node mass due to thyroglossal duct cyst carcinoma is a rare presentation. A 25 year old female presented with a sub mental neck node which was noticed for 6 weeks with no history of fever or any oral lesions. On examination, there was a 1 × 1 cm non tender, firm, mobile sub mental neck node. Fine Needle aspiration cytology (FNAC) of the submental node showed features of metastatic papillary carcinoma from thyroid. Whole body Technitium 99 Pertechnate scan was done to find the primary site and metastases in other neck nodes but failed to find any. After necessary investigations, Total thyroidectomy with sistrunk's procedure, central compartment clearance and level I clearance, was done. Histopathology report showed papillary carcinoma arising from Thyroglossal duct with metastases in submental lymph node. Different pathologies of a mass occurring in submental area are reactive lymphoid hyperplasia, non-Hodgkin lymphoma, dermoid cyst, abscess, sarcoidosis, hemangioma, and lipoma. Neck mass in submental area occurring due to metastases from thyroglossal duct cyst carcinoma without the swelling in primary is a rare presentation.

**Keywords:** Papillary carcinoma, Thyroglossal cyst, Sistrunk procedure, Submental neck node

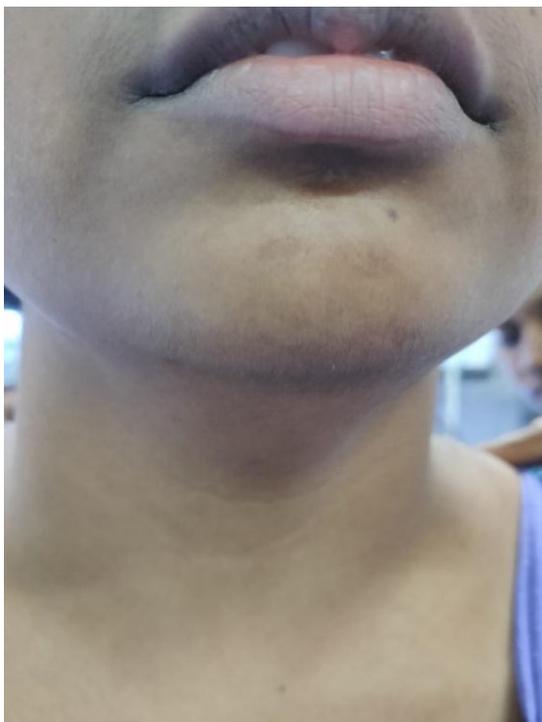
### INTRODUCTION

Neck swelling or neck node in sub mental area is not uncommon. It occurs mostly due to infective or malignant pathologies in oral cavity. Submental neck node mass due to thyroglossal duct cyst carcinoma is extremely rare presentation. Thyroglossal duct carcinomas (TGDC) are also rare, with less than 300 reported cases since the first report in 1915.<sup>1</sup> Thyroglossal duct cysts are the most common anomaly in thyroid development. They are second only to enlarged cervical lymph nodes as the cause of neck mass. Generally, duct cysts are benign, but 1% of cases may be malignant.<sup>2</sup> Thyroglossal cyst can present as a mass located: (a) between the hyoid bone and the thyroid

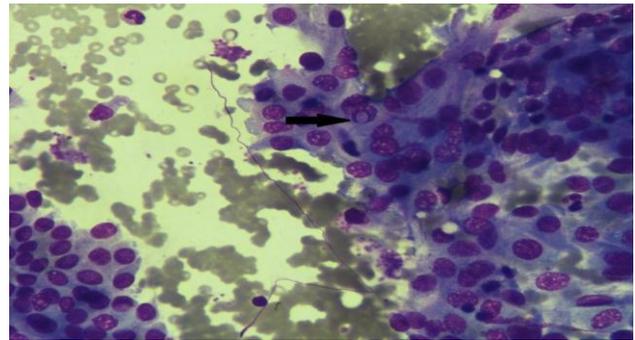
cartilage-60%, (b) above the hyoid bone including the submental site-24%, (c) substernal region-13% and (d) intra-lingual-2%.<sup>3,4</sup> In a review of literature by Weiss et al, out of 115 cases of TGDC, the different types of neoplasia of thyroglossal duct described included thyroid papillary carcinoma in 81.7 per cent, mixed papillary-follicular carcinoma in 6.9 per cent, squamous cell carcinoma in 5.2 per cent, follicular and adenocarcinoma in 1.7 per cent each, and malignant struma, epidermoid carcinoma and anaplastic carcinoma in 0.9 per cent each.<sup>5</sup> Our case of submental neck swelling as a sole presentation which turned out to be metastatic papillary carcinoma from thyroglossal duct is an extremely rare presentation.

## CASE REPORT

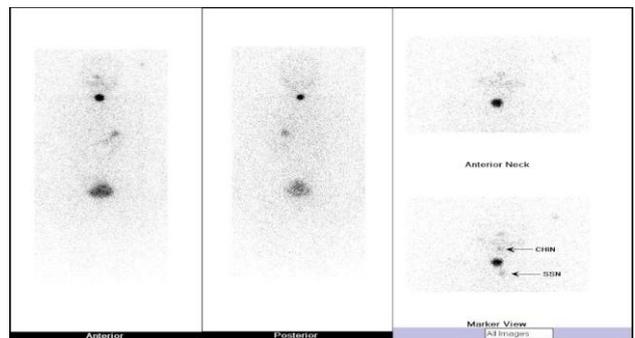
A 25 year old female presented with a sub mental neck node which was noticed for 6 weeks with no history of fever or any oral lesions (Figure 1). The node was not associated with pain, variation in temperature or sudden increase in size. There was no history of dysphagia, dyspnea or hoarseness of voice. On examination, the node was 1 × 1 cm size and found to be non-tender, firm, not moving on swallowing, with no other neck swelling palpable. Ultrasound of neck showed 2 enlarged level IA lymph nodes with loss of fatty hilum within and size measuring about 14 × 8 mm and 13 × 9 mm. Also tiny hypoechoic nodules less than 3 mm were noted in both lobes of thyroid, size of both lobes of thyroid and isthmus measuring normal. Fine needle aspiration cytology (FNAC) of the sub mental neck swelling showed features suggestive of papillary carcinoma (Figure 2). The patient underwent Tc 99 Pertechnate scan in order to find the primary site and to find out the possibilities of metastases in other neck nodes. There was heterogeneous tracer uptake in the sub-mental region and normal uptake in thyroid gland, but no uptake in thyroglossal duct region (Figure 3). Patient's thyroid profile was normal. We performed total thyroidectomy with Sistrunk's operation with clearance of central compartment and level I. Intraoperative and post-operative period was uneventful. Histopathology report showed papillary carcinoma of thyroglossal duct region with metastatic carcinoma in level IA node and thyroid gland showing features of nodular goiter with autoimmune thyroiditis (Figure 4). The patient is under follow up for radioactive iodine treatment as the tumor was extending close to margins. We intend to publish this case report as it is a rare case.



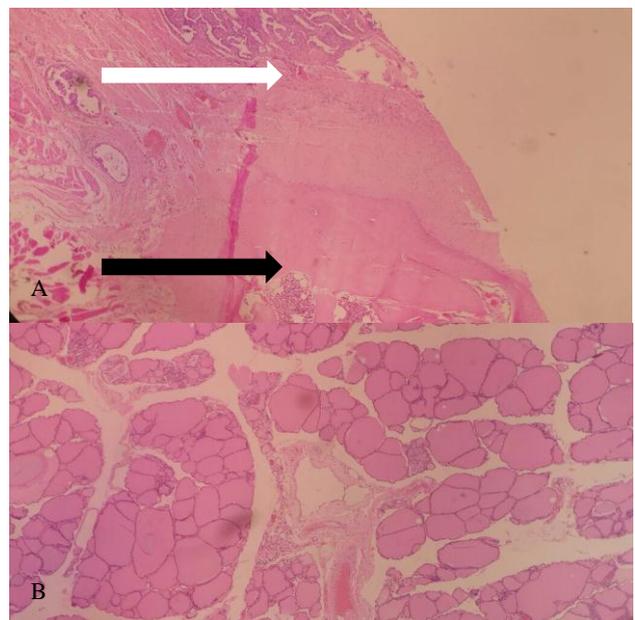
**Figure 1: Patient with submental swelling.**



**Figure 2: FNAC of submental neck node showing intranuclear vacuole (black arrow) feature suggestive of papillary carcinoma.**



**Figure 3: Tc 99 Pertechnate scan showing heterogeneous tracer distribution in the thyroid gland and focal tracer concentration seen in level IA suggestive of nodal deposit.**



**Figure 4: (A) Histopathological examination showing features of papillary carcinoma (white arrow) along with bone (black arrow) and skeletal muscle from thyroglossal duct and (B) Features of nodular goiter in thyroid gland.**

## DISCUSSION

According to a study done by Ural et al, different pathologies of a mass occurring in submental area are reactive lymphoid hyperplasia, non-Hodgkin lymphoma, dermoid cyst, abscess, sarcoidosis, hemangioma, and lipoma.<sup>5</sup> Neck mass in submental area occurring due to metastases from TGDC without the swelling in primary is an extremely rare presentation. The thyroid gland descends from the foramen caecum to its location at the point below the thyroid cartilage. It leaves behind an epithelial tract known as the thyroglossal tract; this tract usually disappears during the 8–10th gestational weeks. Incomplete atrophy of the thyroglossal tract or retained epithelial cysts, create the basis for the origin of a thyroglossal duct cyst.<sup>6,7</sup> The histologic findings of TGDC are most commonly papillary carcinoma (75–80%), but other thyroid tumors such as follicular, Hürthle cell, and mixed papillary–follicular carcinomas have been reported.<sup>8</sup> Imaging diagnostic techniques, including ultrasound, scintigraphy and CT, are usually unable to preoperatively diagnose malignant disease and fine needle aspiration yields a correct result in only 66% of the cases. Nearly 4% of TGDC were found to be locally invasive, while 11% were found to include metastasis to cervical lymph nodes. In about 20% of TGDC, carcinoma in thyroid co-existed.<sup>1</sup> The etiology of the papillary carcinoma arising in a thyroglossal duct is unclear. Generally, there are two theories which can explain this phenomenon, de novo origin and spread from a primary thyroid gland tumor. Most authors support the theory of primary de novo origin by the ectopic thyroid nests of the cyst wall rather than the metastatic spread from a primary thyroid gland tumor through the duct from the thyroid carcinoma, which explains our case. The other theory explains synchronous occurrence of thyroglossal duct cyst carcinoma and thyroid carcinoma, which are reported to be even rarer, as multifocal tumor.<sup>9</sup> Multicentricity and multifocal growth are common in papillary thyroglossal duct carcinomas, as a second malignant lesion is present in 10% of cases with thyroid cancer.<sup>3</sup> In case of large tumors >1 cm, or invasion through the duct cyst wall or suspect foci in the thyroid gland, or a metastases in neck node, a total thyroidectomy followed by I<sup>131</sup> ablation and thyroid-stimulating hormone suppression is the most frequently proposed treatment.<sup>10</sup>

## ACKNOWLEDGEMENTS

We acknowledge Apollo hospitals for using the medical records.

*Funding: No funding sources*

*Conflict of interest: None declared*

*Ethical approval: Not required*

## REFERENCES

1. Yang SI, Park KK, Kim JH. Papillary carcinoma arising from thyroglossal duct cyst with thyroid and lateral neck metastasis. *Int J Surg Case Rep.* 2013;4(8):704-7.
2. Gupta N, Dass A, Bhutani M, Singhal SK, Verma H, Bhutani M, et al. Papillary carcinoma in thyroglossal duct cyst: An unusual case. *Egypt J Ear Nose Throat Allied Sci.* 2014;15:45–7.
3. Doshi SV, Cruz RM, Hilsinger RL. Thyroglossal duct carcinoma: a large case series. *Ann Otol Rhinol Laryngol.* 2001;110(8):734-8.
4. Tradati N, DePaoli F, Benazzo M, Andriele J, Calabrese L, Giugliano G, et al. Papillary carcinoma in thyroglossal duct remnants presentation of four cases and decision procedure for prophylactic thyroid gland dissection. *Oncol Rep.* 2000;7(6):1349-53.
5. Ural A, Imamoğlu M, Umit Işık A, Bahadır O, Bektaş D, Cobanoğlu B. et al. Neck masses confined to the submental space: our experience with 24 cases. *Ear Nose Throat J.* 2011;90(11):538-40.
6. Weiss SD, Orlich CC. Primary papillary carcinoma of a thyroglossal duct cyst: report of a case and literature review. *Br J Surg.* 1991;78(1):87-9.
7. Wexler MJ. Surgical management of thyroglossal duct carcinoma: is an aggressive approach justified? *Can J Surg.* 1996;39(4):263-4.
8. Van Vuuren PA, Balm AJ, Gregor RT, Hilgers FJ, Loftus BM, Delprat CC, et al. Carcinoma arising in thyroglossal remnants. *Clin Otolaryngol Allied Sci.* 1994;19(6):509-15.
9. Sobri FB, Ramli M, Sari UN, Umar M, Mudrick DK. Papillary Carcinoma Occurrence in a Thyroglossal Duct Cyst with Synchronous Papillary Thyroid Carcinoma without Cervical Lymph Node Metastasis: Two-Cases Report. *Case Rep Surg.* 2015;2015:872054.
10. Kristensen S, Juul A, Moesner J. Thyroglossal cyst carcinoma. *J Laryngol Otol.* 1984;98:1277–80.

**Cite this article as:** Manohar B, Abdurahiman R. Thyroglossal duct papillary carcinoma presenting as submental neck node. *Int J Otorhinolaryngol Head Neck Surg* 2019;5:1745-7.