JCHS
e-ISSN: 0128-1089

Available online at
https://jchs-medicine.uitm.edu.my

Journal of Clinical & Health Sciences 10 (2) 2025, 73– 77

Journal of Clinical & Health Sciences

CASE REPORT

Thyroglossal Duct Carcinoma: A Rare Entity

Eric Tan Jin Wee[1,2], Sharir Asrul Asnawi[2], Irfan Mohamad[1*]

[1]*Department of Otorhinolaryngology-Head and Neck Surgery, School of Medical Sciences, Health Campus, Universiti Sains Malaysia*
[2]*Department of Otorhinolaryngology, Hospital Sultanah Nora Ismail, Johor, Malaysia*

Corresponding author: Irfan Mohamad (irfankb@usm.my)

ARTICLE INFO

Article history:
Received
21 February 2024
Revised in revised form
28 May 2024
Accepted
26 June 2024
Published
1 September 2025

Keywords:
thyroglossal duct cyst, papillary carcinoma, Sistrunk procedure

DOI:
10.24191/jchs.v10i2.8584

ABSTRACT

Thyroglossal dust cysts are commonly located over the neck centrally along the path of migration of the thyroid gland. The occurrence of malignancy within thyroglossal dust cysts is extremely rare with papillary carcinoma as the most common subtype. We highlight a case of a 30-year-old gentleman who presented with anterior neck swelling for a month which gradually increased in size. Subsequently post Sistrunk procedure with thyroid gland preservation, he was diagnosed with papillary carcinoma within a thyroglossal dust cyst. He recovered well with no evidence of recurrence with normal blood parameters after one year of follow-up. Although thyroglossal dust cysts malignancy is rare, every patient should be examined thoroughly for risk stratification for malignancy. Any swellings over neck region is warranted for a proper evaluation by the attending clinician.

INTRODUCTION

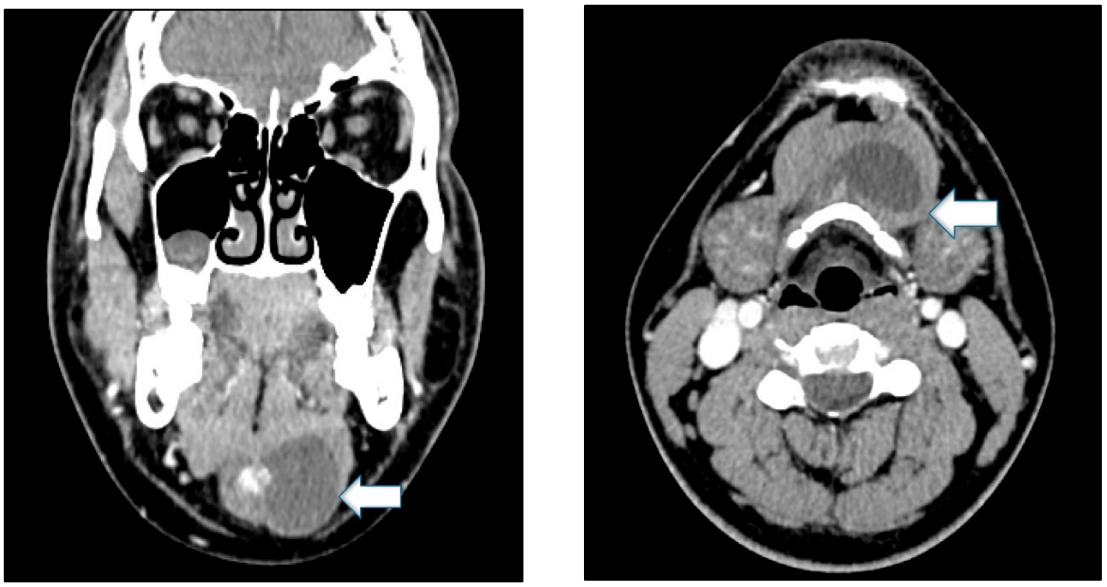
The thyroid gland descends from the foramen cecum around the third week of fetal life. An epithelial tract known as the thyroglossal duct is left behind which usually atrophies and disappears between 7th and 10th weeks of gestation [1]. The thyroglossal duct does not undergo complete involution in some individuals thus creating a duct, cyst or fistula anywhere along the tract.

Thyroglossal duct cyst (TDC) are the most common anomaly in thyroid development. It contributes around 70% of midline masses presenting in childhood and 7% in adulthood [2]. Carcinoma arising from a TDC is a rare complication affecting less than 1% of cases with majority of the primary carcinoma to be papillary carcinoma followed by follicular carcinoma [2]. In addition, no cases of medullary thyroid carcinoma had been reported arising from TDC in view of parafollicular or C cells arises from a different embryological origin. The current consensus defined TDC carcinoma as a neoplasm arising "de novo" within a TDC and not metastasised from the thyroid gland. Most cases of TDC carcinoma are usually asymptomatic and often incidentally diagnosed via histopathological examination after surgical removal from a presumed case of TDC.

CASE PRESENTATION

A 30-year-old gentleman was referred with a chief complain of a painless midline neck swelling for the past one month, which has been gradually increasing in size with no skin changes. He has no known medical illness or any significant family history suggesting malignancy. Furthermore, he has no past surgical history. Physical examination showed a well-demarcated, firm and non-tender midline swelling above the hyoid bone of about 4 centimeters in diameter. There were no areas of fluctuant or tenderness upon palpation and the swelling moved with protrusion of his tongue. The thyroid gland was otherwise unremarkable and there were no enlarged cervical lymph nodes.

Full blood count and thyroid function tests were normal. Ultrasonography (USG) showed a cystic lesion with debris within measuring 3.0 X 3.1 X 2.8 cm (APxWxCC). The thyroid gland was homogenous with no focal lesion and no cervical lymphadenopathy. To further evaluate the swelling, a computed tomography (CT) imaging scan was done (Figure 1). Fine-needle aspiration (FNA) and cytology of the lesion yield 5ml of straw-coloured yellowish fluid that showed atypical cells arranged in cohesive clusters and papillary like structures exhibiting nuclear pleomorphism that were suspicious of malignancy.



(a) (b)

Figure 1 (a) Well defined non-enhancing cystic lesion located at the left sublingual region (arrow). No areas of microcalcifications or necrosis seen. (b) well circumscribed hypodense lesion with cystic attenuation located at the left paramidline and level of the hyoid bone (arrow).

As there was suspicion of malignancy arising from the lesion, he underwent primary Sistrunk procedure without a total thyroidectomy (Figure 2) as he carried a low risk of developing TDC carcinoma which was further supported by the neck USG. The surgery procedure was uneventful. He was discharged on day two postoperatively with no complications. The histopathology of the excision specimen showed a maximum diameter of 3.7 cm with papillary architecture lined by columnar cells with pleomorphic elongated to oval nuclei invading the cyst wall suggesting of papillary thyroid carcinoma. All margins were clear with the nearest surrounding margin of 5mm (Figure 3). The index patient was closely monitored in the clinic every month for the first six months. Currently, he is having half yearly follow up with no signs of recurrence and normal thyroglobulin level.

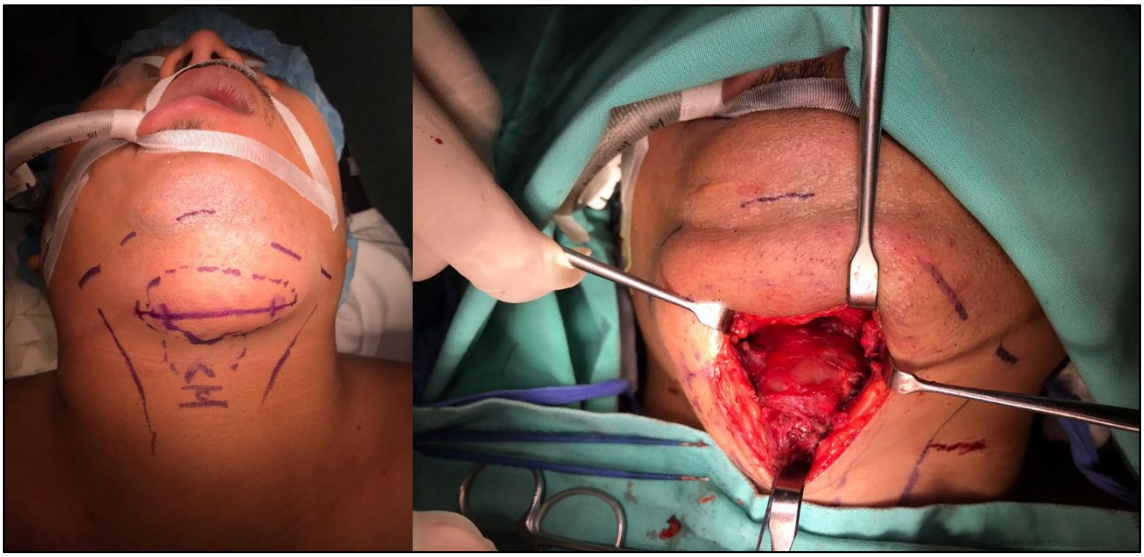


Figure 2 The midline swelling prior to surgery and excision of the thyroglossal duct cyst (Sistrunk procedure)

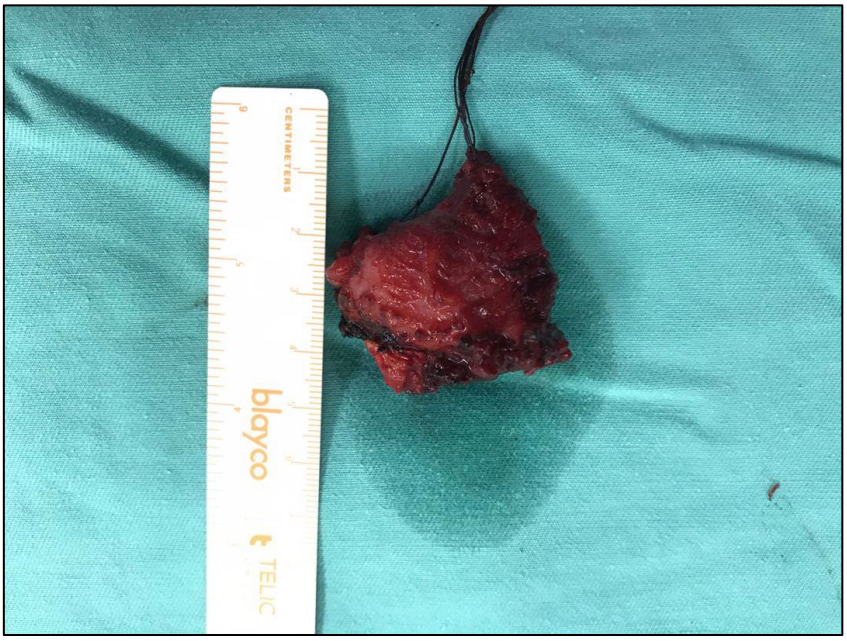


Figure 3 The complete removal of the swelling and tract with a measurement of 4 x 3 cm (width x length)

DISCUSSION

A thyroglossal duct remnant can present as a cyst, sinus, fistula or as an ectopic thyroid with cysts formation being the commonest anomaly. It was first described in 1920, the mainstay of treatment for TDC is the surgical excision of the cyst known as Sistrunk procedure which involves the removal of the entire length of the duct and the central portion of the hyoid bone. In the general population there is around 7% of TDC that failed to close, with a ten-fold increase in the paediatric age group [2].

Complications such as sinus formation and infection are well documented in literatures but malignant changes within TDC is rare with less than 1%. Up to date there are approximately 250 cases published literature with the first case reported in 1911 [3]. The pathophysiology of development of malignancy in a TDC is still unclear but there are two predominating theories; either metastatic disease from an occult primary or spontaneous development from ectopic thyroid tissue within the cyst wall [4]. Initial signs and symptoms of a TDC carcinoma mimic other benign lesions that makes establishing an accurate diagnosis harder. Malignancy from a TDC is more favourable to women with the mean age in the fourth decade. For the index patient he is a male with a young age of 30.

Most cases of TDC carcinomas were papillary carcinoma then follicular carcinoma followed by squamous cell carcinoma carrying a worse prognosis. Overall, three quarter of TDC carcinomas were diagnosed as incidental findings on final pathologic analysis. History, examination findings in addition with imaging, such as USG or CT scan are important workup for TDC. Imaging is always mandatory in order to evaluate the morphology of the nodule and presence of suspicious features such as microcalcification, hypervascularisation, prominent solid part and asymmetrical wall thickening which carry a higher risk of malignancy. Tissue cytology such as FNA with image guided reach values of sensitivity and specificity up to 100%. Due to low cost and high accuracy, FNA is recommended in all patients treating for TDC.

The treatment of TDC carcinoma still remains controversial among surgeons, with the concern of perseveration of the thyroid gland. Sistrunk procedure is the gold standard for removal of TDC in literatures. The next step in management pertains to the thyroid gland. The current theory is that papillary carcinomas arise from the TDC de novo and are not metastatic from the thyroid gland as there are no reports of TDC medullary carcinoma [5]. In general, patients are treated accordingly based on risk assessment of different individuals. Characteristics such as age younger than 45 years with no history of radiation and tumour size of less than 4 centimetre, without extracystic extension and no locoregional and no distant metastasise carry a low risk factor for developing thyroid malignancy [6]. Thus, Sistrunk procedure suffices in cases of TDC with 95-100% long term survival as majority of the patients are from low-risk category. In patients with advanced disease, including spread to cervical lymph nodes or extracapsular invasion into thyroid gland, laryngeal cartilage, and vessels a total thyroidectomy with neck dissection and radioactive iodine ablation is indicated. According to Lancini et al, high risk patients who underwent the mentioned procedures above had been proven to have long term survival rate of more than 95% [7].

From the reported case, we chose Sistrunk procedure with thyroid preservation as the treatment strategy for the patient because he has low-risk based on the risk stratification. Currently, he is having half yearly follow up with Otorhinolaryngology and Breast and Endocrine teams. His latest neck ultrasound and thyroglobulin level were normal. USG with serum monitoring will be done yearly to look for recurrence.

CONCLUSION

TDC carcinoma usually presents similar to their benign counterpart as a benign looking neck swelling which is often only diagnosed after removal post surgically as an incidental finding on histopathological examination. Clinicians should be aware about the incidence of this disease and thorough history and examinations should be done. Surgery remains the mainstay of treatment for TDC. Currently, there are no evidence-based clinical guidelines that have been published on the optimal surgical approach for cases of TDC carcinoma. A multidisciplinary approach should be considered to safely identify high-risk patients who require a more aggressive treatment approach and follow up.

Consent

Informed consent was obtained from the patient for case write up including permission for publication of all photographs and image.

CONFLICT OF INTEREST

The authors agree that this research was conducted in the absence of any self-benefits, commercial or financial conflicts and declare the absence of conflicting interests.

ACKNOWLEDGEMENT

The authors are incredibly grateful to our patient cooperation and permission to present his case to the scientific community. Furthermore, we will like to thank the otorhinolaryngology specialist and other multidisciplinary team members from Hospital Sultanah Nora Ismail who were involved in treating this patient while he was in the ward and when attending out-patient clinic.

AUTHORS' CONTRIBUTIONS

ET and SA had decided and wrote the initial draft of the manuscript. IM provided supervision and edited the manuscript. All author's had contributed significantly in this manuscript.

REFERENCES

1. Ellis PD, van Nostrand AW. The applied anatomy of thyroglossal tract remnants. Laryngoscope 1977; 87: 765e70.

2. Carter Y, Yeutter N, Mazeh H: Thyroglossal duct remnant carcinoma: Beyond the sistrunk procedure. Surg Oncol, 2014; 23(3): 161–166.

3. Chrisoulidou A, Iliadou P, Doumala E, Mathiopoulou L, Boudina M, Alevizaki M, Patakiouta F, Xinou E, Pazaitou-Panayiotou K. Thyroglossal duct cyst carcino¬mas: Is there a need for thyroidectomy? Hormones, 2013; 12(4): 522–528.

4. Underwood HJ, Williams DM, Kundel A: Papillary thyroid carcinoma with¬in thyroglossal duct cyst: Case report and review of literature. JSM Head Neck Cancer Cases Rev, 2016; 1(2): 1006

5. Rayess HM, Monk I, Svider PF, Gupta A, Raza SN, Lin HS. Thyroglossal duct cyst carcinoma: A systematic review of clinical features and outcomes. Otolaryngol Head Neck Surg. 2017; 156(5): 794-802.

6. Wood CB, Bigcas JL, Alava I, Bischoff L, Langerman A, Kim Y. Papillary-type carcinoma of the thyroglossal duct cyst; the case for conservative management. Ann Otol Rhinol Laryngol 2018; 127:710-716.

7. Lancini D, Lombardi D, Piazza C. Evidence and controversies in management of thyroglossal duct cyst carcinoma. Curr Opin Otolaryngol Head Neck Surg. 2021 Apr 1;29(2):113-119.