

Papillary Thyroid Carcinoma of a Large Thyroglossal Duct Cyst: A Case Report

Akoijam Romesh Singh¹, Phungreikan Ningshen², Sanamacha Leishangthem¹, Kongsang SR¹, Ng. Ngajokpa Martin¹, A. Santhosh¹, Sruthy Ashok¹, Nongmeikapam Romesh Singh¹

¹PGT, Department of General Surgery, Jawaharlal Nehru Institute of Medical Sciences (JNIMS), Imphal

²Assistant Professor, Department of General Surgery, Jawaharlal Nehru Institute of Medical Sciences (JNIMS), Imphal

ABSTRACT

Background: Thyroglossal duct cyst is the most common congenital anomaly of the thyroid gland. Carcinoma arising from it is rare which composes 1% of total cases, often diagnosed incidentally after surgical excision. Here we are reporting a case of papillary thyroid carcinoma of thyroglossal cyst in 45-year-old female with normal thyroid gland.

Case Description: A 45-year-old female presented with 1 year history of swelling over midline of the neck with normal thyroid gland. FNAC suggested a benign cystic lesion, most likely thyroglossal cyst. CT scan neck showed a well-defined lobulated and septated thin-walled cystic lesion of approx. 6×4×5cm in the midline upper infrahyoid region anterosuperior to thyroid, suggestive of thyroglossal cyst. Sistrunk procedure was performed. HPE revealed papillary thyroid carcinoma of thyroglossal cyst.

Conclusion: A case of thyroglossal duct cyst with normal thyroid gland and without any clinical or radiological evidence of malignancy, a possibility of underlying malignancy should be kept in mind and can be managed adequately with Sistrunk procedure only, with regular follow up

KEYWORDS: Thyroglossal cyst, papillary thyroid carcinoma, Sistrunk procedure

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INTRODUCTION

During the third week of fetal life the thyroid gland develops from an invagination of the midline of endoderm cells. This epithelial invagination descends from the base of the tongue along the midline towards the anterior section of the neck towards the first and second tracheal rings.^[1] Thyroglossal duct cysts are the most common anomaly in thyroid development.^[2] 70% of midline masses in the neck are thyroglossal cysts in children and in adults they are 7% of midline masses.^[3] Thyroid carcinoma is a rare complication of untreated thyroglossal duct cysts, affecting less than 1% of cysts.^[4,5] Carcinomas arising from it are usually characterized by nonaggressive behaviour with rare lymphatic spread. Most of them are diagnosed during the third and fourth decades of life and rarely in children under 14 years.^[6] The most common primary thyroglossal duct cyst carcinoma is papillary carcinoma (75–80%), other tumour types are mixed papillary-follicular carcinomas (7%), squamous cell carcinoma (5%), follicular carcinoma (1.7%); Hurthle cell carcinoma and anaplastic carcinoma (0.9%) have also been reported.^[7]

Sistrunk's procedure is usually done for thyroglossal duct cyst. Sistrunk's procedure comprises of removal of the thyroglossal duct cyst, the central portion of the body of the hyoid bone, and a core of tissue around the thyroglossal tract to open into the oral cavity at the foramen cecum.^[8] Sistrunk's procedure is commonly performed in low-risk patients. For high-risk patients, they are likely to undergo an additional total thyroidectomy and postoperative radioactive iodine ablation therapy.^[9]

Here we are reporting a case of papillary thyroid carcinoma of thyroglossal duct cyst in 45-year-old female with normal thyroid gland.

CASE REPORT

A 45-year-old female was presented in our Surgery Out Patient Department, Jawaharlal Nehru Institute of Medical Sciences, Manipur, India in May 2023, with 1 year history of swelling over midline of the neck. The swelling was giving a double chin appearance. It was rapidly increasing in size in the later 5 months of the year. The swelling was not associated

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with any pain. It was also not associated with dyspnoea/dysphagia/dysphonia.

On clinical examination a midline infra-hyoid swelling of size 10x10 cm was present and it was extending upto upper border of the thyroid gland (figure.1). The swelling moved with deglutition but not moving with the protrusion of tongue.

Surface of the swelling was smooth and the margin was well defined.

On palpation it was non tender and firm in consistency. It was of limited mobility on both vertical and horizontal directions. There were no palpable cervical lymph nodes.

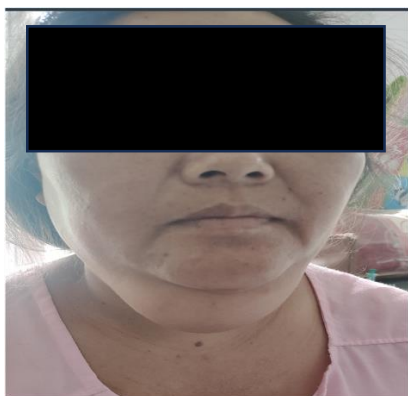


Figure.1

Fine needle aspiration cytology was sent for the neck swelling and it was suggestive of a benign cystic lesion, most likely thyroglossal cyst. Computed tomography of the neck showed a well-defined lobulated and septated thin-walled cystic lesion of approx. 6x4x5cm in the midline upper infrahyoid

region anterosuperior to thyroid which was suggestive of thyroglossal cyst and with a right colloid nodule of size 0.8cm (figure.2).

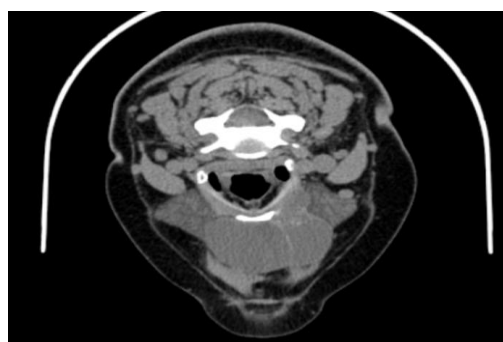
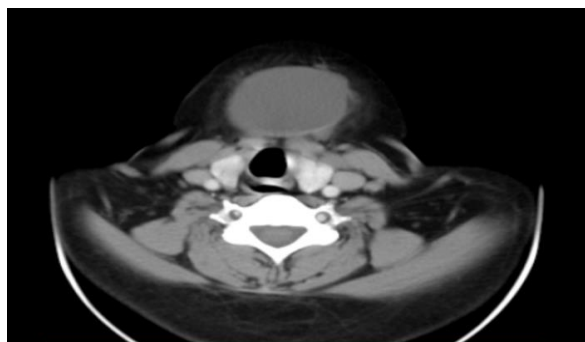


Figure.2 CT Scan Neck

Thyroid function tests were within normal limits. Haematological and biochemical investigations turned out to be normal. Patient was admitted and was prepared for surgery. Sistrunk's operation was done (figure.3).

Intra-operatively there was a cystic swelling of size 6x6 cm, adherent to platysma and intra-cystic septations along with a nodule 0.5x0.5 cm.

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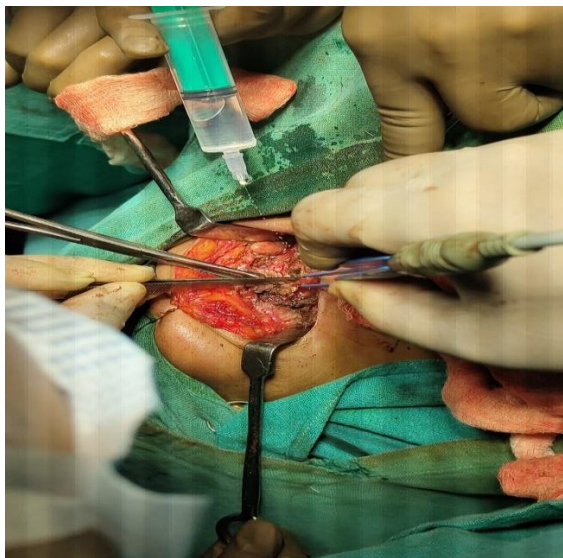


Figure 3. Sistrunk's Operation



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Histopathological examination was done for the excised thyroglossal duct cyst and serial sections from the nodules showed numerous arborising capillaries with well-formed fibrovascular cores lined by thyroid follicular epithelial cell. Many of the cells display longitudinal nuclear grooves and

note is made of an occasional nuclear pseudo-inclusion. Focal nuclear overcrowding was noted. Some scattered psammoma bodies were also seen. Features were suggestive of papillary thyroid carcinoma (figure.4).

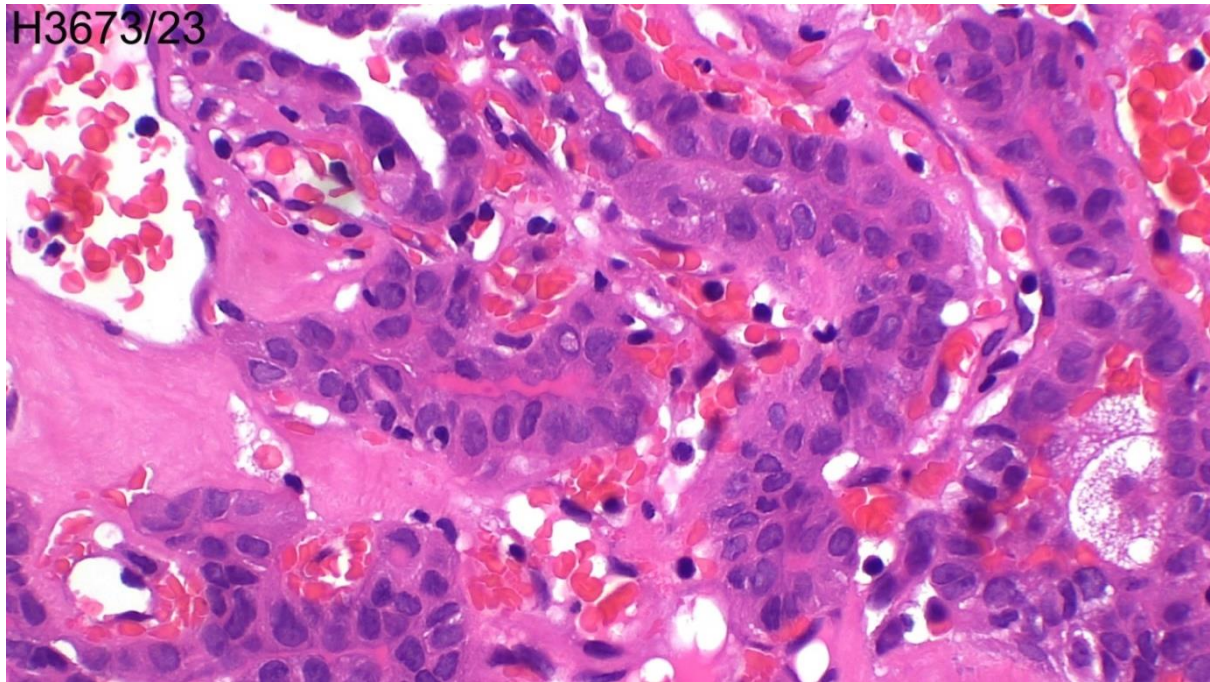


Figure 4. HPE suggestive of papillary Thyroid Carcinoma of Thyroglossal Cyst

Post operative period was uneventful and patient was discharged after 5 days post-surgery (figure.5). Currently, at

8 months of follow-up, the patient remains free from recurrent thyroid tumour.



Figure 5. Post-operative pictures

DISCUSSION

Papillary carcinoma originating from a thyroglossal cyst represents an infrequent finding, which occurs in approximately 1% of thyroglossal cyst cases. They usually present the same as that of a benign thyroglossal cyst, which represents the most frequent benign congenital lesion of the

neck. This type of cancer is more common in adults with a 2.3:1 female:male ratio.^[7] The mean age of patients is in the fourth decade of life and the tumour can be located anywhere along the embryological route of descent of the thyroid gland.^[10]

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In this case report, the patient was a 45-year-old woman who was same as the mean age of presentation for this tumour and with a midline neck mass gradually increased during the next 5 months. The rate of co-existence of thyroglossal duct cyst and thyroid carcinoma is between 0–25%.^[11] The pre-operative evaluation of patients who are discovered to have a thyroglossal duct cyst should include a complete physical examination, accurate head and neck examination, palpation of the thyroid gland, thyroid function tests and a thyroid scan.^[12] In the present case, thorough preoperative evaluation, as well as pathological examination of the thyroid were done. The thyroid function tests were normal.

Malignancy in thyroglossal duct cyst can be suspected when the swelling is not moving freely on deglutition along with dysphasia, dysphonia and lymphadenopathy. However, in our case, movement with deglutition and tongue protrusion could not be appreciated due to large swelling size and short neck. Investigations like ultrasound and computed tomography do not allow a preoperative diagnosis, and fine-needle aspiration cytology yields a correct result in only 66% of the cases.^[13] Sistrunk procedure is commonly done for thyroglossal duct remnants. We removed the thyroglossal duct, including the cyst, the middle part of the hyoid bone and the rest of the duct extending to the base of the tongue. 62 cases of these carcinomas was analysed by Patel et al. and suggested that Sistrunk procedure is adequate for most patients and has low rates of recurrence (1.8%).^[14] Papillary carcinoma is the most common malignancy found in a thyroglossal duct cyst (80%), followed by follicular or mixed papillary-follicular carcinoma (9%), squamous cell carcinoma (5%), adenocarcinoma (2%), anaplastic carcinoma (1%) and others (3%).^[15] The prognosis of papillary carcinoma arising in thyroglossal duct cyst is good and has an overall survival rate of 95.6% at 10 years.^[16] According to the Widstrom et al. criteria, the diagnosis of primary carcinoma of the thyroglossal duct includes the histological identification of thyroglossal duct carcinoma, demonstration of the normal epithelial lining of the thyroglossal duct, normal thyroid follicles within walls of the cyst, normal thyroid tissue adjacent to the tumour and no findings of primary thyroid carcinoma on histopathological examination of the thyroid gland.^[17] Microscopic examination of the specimen in this presented case suggested of primary papillary carcinoma arising in a thyroglossal duct cyst.

The prognosis of papillary thyroid carcinoma of thyroglossal duct cyst is very good and metastatic spread is not very common.^[18,19] In a study by Patel et al., with a median follow-up of 71 months, the 5-year and 10-year Kaplan-Meier overall survival (OS) was 100% and 95.6%, respectively.^[20]

Papillary carcinoma is usually a low-grade malignancy and careful long-term follow-up is important. Recurrences if present can be successfully treated with careful patient follow-up.^[21] During follow-up patients must have a neck scan and should be re-assessed every six months during the

first year and annually after that.^[22] Follow-up procedures consist of physical examination, ultrasound of the surgical region, and thyroid and total body scintigraphy as proposed by Park.^[23]

CONCLUSION

Thyroglossal duct cyst carcinoma is a rare tumour and it is often diagnosed postoperatively as an incidental finding on histopathological examination. Surgery remains the cornerstone of treatment. There are no evidence-based clinical guidelines that have been established on the optimal surgical approach and further management at present. A multidisciplinary approach should be considered to safely identify high-risk patients, who will require a more aggressive treatment approach. A possibility of underlying malignancy should be kept in mind, for a case of large thyroglossal duct cyst with normal thyroid gland, which has no clinical or radiological evidence of malignancy. It can be managed adequately with surgical procedure only followed by a regular follow up.

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