

Papillary carcinoma of thyroid in a thyroglossal cyst

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Abstract

Thyroglossal duct cyst (TDC) is the most common congenital anomaly of the thyroid gland and the most common congenital cervical abnormality in childhood. Carcinoma arising from a TDC is rare, which composes only 1% of TDC cases. Here, we are reporting a case of 50-year-old male with a swelling in the upper left neck region which was progressively increasing in size over 5 years. Ultrasonography report showed a large lobulated cystic mass. Internal echoes with calcification and there were no neck nodes. Thyroid gland was normal. In the present study, the Sistrunk procedure was used to excise a TDC. Papillary carcinoma was confirmed with the histopathological examination following the surgery. The patient was treated with radioactive iodide and thyroid suppression therapy was given as adjuvant treatment. The patient has been following for 2 years without any metastasis.

Keywords: Papillary carcinoma, sistrunk procedure, thyroglossal duct cyst, ultrasonography

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INTRODUCTION

Thyroglossal duct cyst (TDC) is the most common congenital anomaly of the thyroid gland as well as midline neck mass.^[1] Normal thyroid gland descends along thyroglossal duct, embryonic midline duct originated from foramen cecum through the floor of mouth and passes anteriorly to inferior part of the neck where thyroid gland is found in adult. TDC is usually located in the midline or, less frequently, in the anterolateral part of the neck (25%).^[2] A failure of regression of the thyroglossal duct may cause cysts of the TDC.^[3] Carcinomas arising from a TDC are rare and are usually characterized by nonaggressive behavior and rare lymphatic spread. Most cases of TDC carcinoma are diagnosed during the third and fourth decades of life and rarely in children under 14 year.^[4] Here,

we are reporting a case of papillary carcinoma of thyroid in a thyroglossal cyst.

CASE REPORT

A 50-year-old male patient came to our ENT outpatient department with complaint of the left side neck swelling which was progressively increasing in size for the past 5 years. Patient was apparently all right 5 years back. Then, he noticed a swelling in the left side of the neck which was slowly increased to present size. Patient had no history of pain, dysphasia, hoarseness of voice, fever, loss of appetite and loss of weight. Patient had no history of any systemic illness and family history. On inspection, a well-defined swelling seen on the left side of the neck measuring approximately 10 cm × 6 cm in dimensions.

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The overlying and surrounding skin appears to be normal. There was no presence of sinus or fistula. On palpation, all inspector findings were confirmed. The swelling was cystic, nonlobulated, nontender predominantly involving the left upper neck and crossing midline. It was fixed to the underlying structure but not to the overlying structures which was compressible, nonpulsatile and nonfluctuant. Movement with deglutition and tongue protrusion could not be appreciated due to large swelling size, short neck and obesity. Thyroid gland was normal by palpation [Figure 1]. No neck nodes were palpable. On the basis of all above findings, the provisional diagnosis was given as thyroglossal cyst and enlisted the differential diagnosis as dermoid cyst, lymphangioma, hemangioma, branchial cyst and lymphoma. The patient was then subjected to further investigations. There was no abnormality detected in routine blood and urine investigations. Fine-needle aspiration cytology was suggestive of cystic swelling. There was no abnormality detected on X-ray chest posteroanterior view; however, the X-ray neck lat view showed a soft-tissue swelling in upper neck [Figure 2]. Ultrasound scan showed an evidence of large lobulated cystic mass noted in the left submandibular region. Fluid shows internal echoes and calcification within [Figure 3]. After thorough clinical examinations and investigations, excision of cystic swelling under general anesthetic was planned. Intraoperative swelling was found to be attached with hyoid bone. Hence, Sistrunk's procedure was performed. Adjacent lymph nodes were removed. Intraoperative diagnosis was TDC [Figure 4a and b]. Specimen was sent for histopathological examination. Gross specimen received was a paramedian neck swelling measuring 11.3 cm × 6 cm × 4.9 cm. External surface is multinodular and well capsulated. Cut section is multilocular, cystic and filled with yellow-brown fluid.

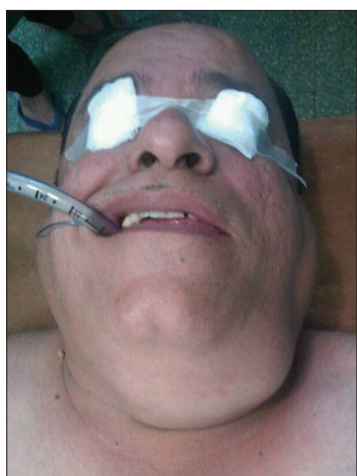


Figure 1: A cystic, nonlobulated swelling predominantly involving the left upper neck and crossing midline

The solid areas were gray yellow with the areas of calcification and focal papillary excrescence measuring 2.2 cm × 1.6 cm × 1 cm [Figure 5]. Microscopic examination showed a multiloculated cyst focally lined by pseudostratified ciliated epithelium. The wall shows thyroid follicles, lymphoid aggregates and foci of cholesterol clefts and calcification. Secondary inflammation is also noted. In areas, the cyst wall shows papillary carcinoma of thyroid exhibiting nuclear grooves and overlapping. The tumor infiltrates the cyst wall reaching close (1 mm) to the inked surgical margin. Adjacent two lymph nodes show reactive hyperplasia [Figure 6a and b]. On the basis of microscopic examinations, the following histopathology impression was made: (i) papillary carcinoma of thyroid in a thyroglossal cyst (ii) the tumor reaches close to the inked surgical margin and (iii) two adjacent lymph nodes free of tumor. Postoperatively, the patient was absolutely alright and symptomatically relieved [Figure 7]. The regular follow-up was done for 2 years.

DISCUSSION

A mass in the neck is a common clinical finding and differential diagnosis may be extremely broad. Most of these masses are due to benign processes, malignant diseases must not be overlooked. Hence, it is important to develop a systematic approach for the diagnosis and management of neck masses. TGD cysts are the most common midline mass found in children. Most of the thyroglossal duct remnants are benign; however, a small percentage of them undergoes neoplastic change.^[5] TDC Carcinoma is uncommon and the incidence varies from 0.7 to 1% of TDC.^[6] It affects women and men at a ratio of 2:1.^[7]



Figure 2: X-ray neck lateral view showed a soft-tissue swelling in the upper neck

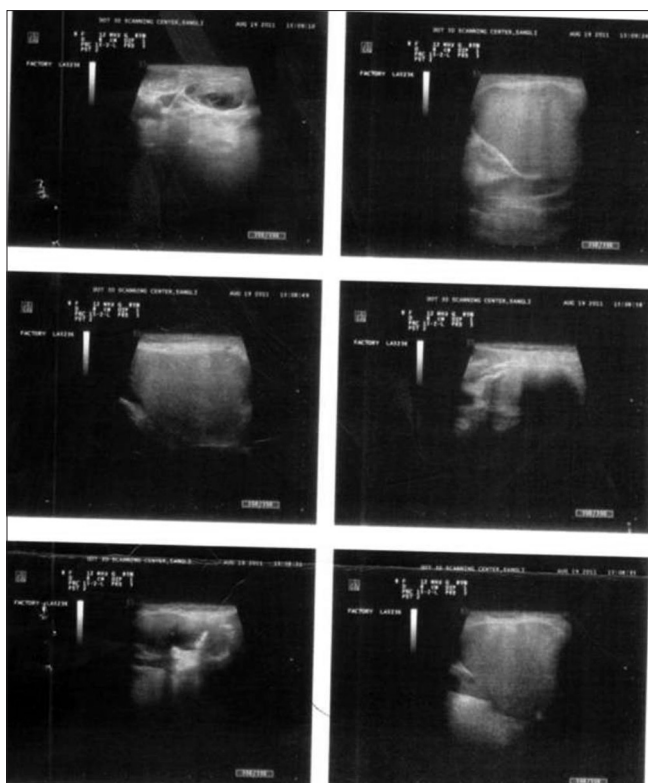


Figure 3: Ultrasound scan showed an evidence of large lobulated cystic mass noted in the left submandibular region. Fluid shows internal echoes and calcification within



Figure 5: Gross specimen received was a paramedian neck swelling measuring 11.3 cm × 6 cm × 4.9 cm. External surface is multinodular and well capsulated. Cut section is multilocular, cystic and filled with yellow-brown fluid. The solid areas are gray yellow with areas of calcification and focal papillary excrescence measuring 2.2 cm × 1.6 cm × 1 cm

If the mass is not moving freely on deglutition and having dysphasia, dysphonia and lymphadenopathy then malignancy in TDS can be suspected. However, in our case, movement with deglutition and tongue protrusion could not be appreciated due to large swelling size, short neck and obesity. Imaging tests (ultrasound and computed

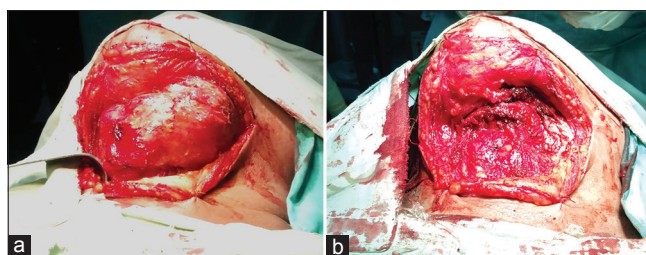


Figure 4: (a and b) Sistrunk's procedure (intermediate pictures)

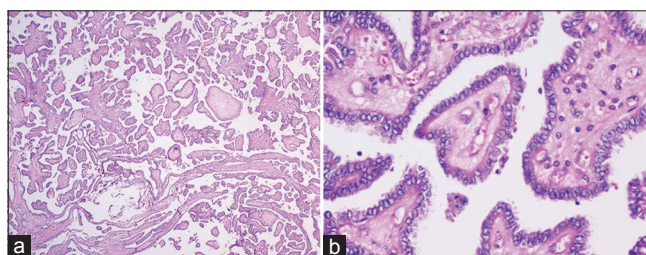


Figure 6: (a and b) Microscopic examination showed a multiloculated cyst focally lined by pseudostratified ciliated epithelium. The wall shows thyroid follicles, lymphoid aggregates and foci of cholesterol clefts and calcification. In areas, the cyst wall shows papillary carcinoma of thyroid exhibiting nuclear grooves and overlapping

tomography) do not allow a preoperative diagnosis, and fine-needle aspiration yields a correct result in only 66% of the cases.^[8]

The surgical excision of a thyroglossal duct remnant consists in a Sistrunk procedure. 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. Adjacent lymph nodes were removed. Patel *et al.* analyzed 62 cases of these carcinomas and suggested that Sistrunk procedure is adequate for most patients and has low rates of recurrence (1.8%).^[9]

Papillary carcinoma is the most common malignancy found in a TDC (80%), followed by follicular or mixed papillary-follicular carcinoma (9%), squamous cell carcinoma (5%), adenocarcinoma (2%), anaplastic carcinoma (1%) and others (3%).^[10]

The prognosis of papillary carcinoma arising in TDC is excellent, with an overall survival rate of 95.6% at 10 years.^[11] The postsurgical follow-up of patients is limited to an annual clinical and ultrasound cervical examination and measurement of thyroid stimulation hormone level.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal.



Figure 7: Postoperative follow-up

The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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