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Case Report

Ear, Nose and Throat Section

Papillary Carcinoma of Thyroid in Thyroglossal Cyst- A Case Report

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ABSTRACT

Thyroglossal duct cysts are most commonly occurring congentinal midline swellling of the neck. Usually carcinomas in thyroglossal duct cysts is extremely rare, commonly known as papillary carcinomas. However, the diagnosis is only made postoperatively after excision of the cyst. Although the Sistrunk procedure is often regarded as adequate but controversies exist, the need for thyroidectomy is based on histopathological findings. This is a case report of 43-year-old male presenting with swelling in the midline of the neck for one year. On examination, a cystic swelling was present in the midline of the anterior aspect of the neck. Ultrasonography (USG) neck revealed heteroechoic cystic lesion with solid component and microcalcifications present within the cyst suggestive of thyroglossal duct cyst probably neoplastic. The mass was surgically excised and sent for histopathological examination and reported as thyroglossal duct cyst with papillary carcinoma of thyroid. Appropriate history, clinical examination and investigation leads to the correct diagnosis and treatment. Incomplete removal of the mass leads to recurrence. Histopathological examination is a must postoperatively. The patient is still on follow-up and no recurrence have been noted.

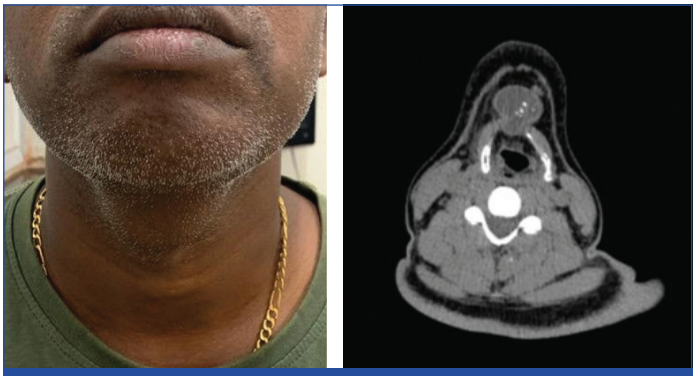
Keywords: Congenital anomaly, Histopathology, Midline neck swelling, Thyroid carcinoma, Thyroid gland

CASE REPORT

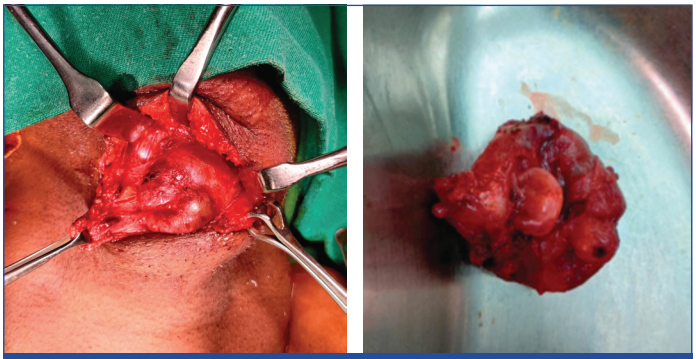
A 43-year-old male presented with the complaint of swelling in the midline of the neck for a duration of one year. The swelling was insidious at onset, gradually progressing and attained the current size. There was no history of associated pain, trauma, etc. No previous history of surgery was recorded. Local examination of ear, nose and throat was found to be normal, however when it was done for neck, it showed a cystic swelling of size approximately measuring 2.5×2 cm present in the anterior aspect of neck 3 cm below the chin, 4.5 cm above the suprasternal notch and 1.5 cm from anterior border of sternocleidomastoid muscle on both sides [Table/Fig-1]. Swelling was ovoid in shape, cystic in consistency, compressible, no warmth, no tenderness. Swelling was mobile on protrusion of tongue. On palpation trachea was in midline. Bilateral carotid pulsations were felt. No cervical lymphadenopathy was seen.

With the above history and clinical examination, patient was investigated further with USG neck, Contrast Enhanced Computed Tomography (CECT) neck and Fine Needle Aspiration Cytology (FNAC). USG Neck- Heteroechoic cystic lesion was seen with solid component and microcalcifications present within the cyst suggestive of thyroglossal duct cyst, which was probably neoplastic. On CECT Neck- Well-defined cyst with peripheral wall enhancement with enhancing solid was noticed within it. Foci of pure tone calcification noted with intralaryngeal extension through anterior hyoid [Table/Fig-2]. In FNAC, numerous macrophages and squamous epithelial cells with colloids were seen. These features were suggestive of possibility of thyroglossal duct cyst.

Under general anaesthesia, orotracheal intubation, patient in supine position with neck extended and sand bag under shoulders, parts painted and draped, local infiltration given with 2% xylocaine and adrenaline. Horizontal skin crease incision was made over the swelling from anterior border of right sternocleidomastoid to the left. Incision was deepened and flaps were elevated in subplatysmal plane till the hyoid bone superiorly and tracheal cartilage inferiorly. Strap muscles were divided in midline and retracted laterally. Cystic swelling,ovoid in shape was visualised [Table/Fig-3]. Swelling was not fixed inferiorly and was separated from the underlying structures. Tract was traced till authors reached hyoid bone. Swelling mobilised inferiorly and separated from the underlying structures. Tract was traced and hyoid bone reached. Muscles attached to the body of the hyoid bone were released and its central part were transected to trace tract superiorly.High ligation was done and specimen was removed [Table/Fig-4]. Haemostasis secured. Suction drain of size 22 and stay sutures were placed. Strap muscles were approximated and midline subcutaneous sutures with subcuticular skin sutures were given. Patient was extubated and shifted to to intensive care unit under stable condition.



[Table/Fig-1]: Preoperative picture of the swelling in midline of neck; **[Table/Fig-2]:** CECT image showing cystic lesion with solid area component and foci of calcification. (Images from left to right)

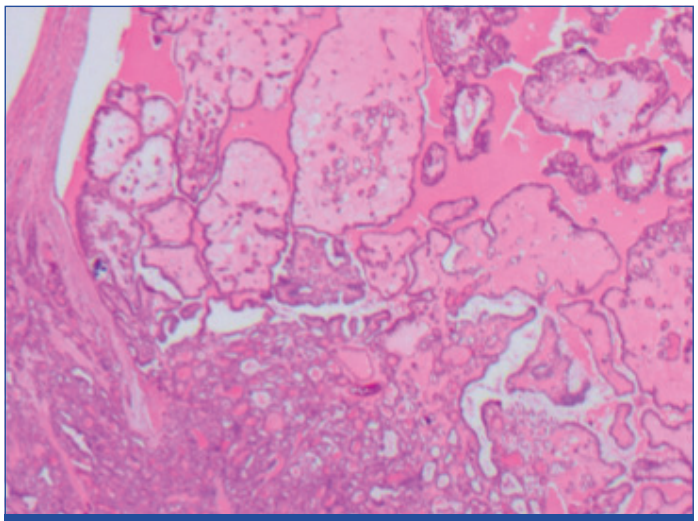


[Table/Fig-3]: Intraoperative image of the cystic swelling; **[Table/Fig-4]:** Gross Specimen: Cystic swelling removed with portion of hyoid bone. (Images from left to right)

Histopathology

Histopathological report showed multifocal tumour in the cystic wall as well as intracystic area. The tumour was composed of thyroid follicular cells, arranged in papillary and follicular patterns [Table/Fig-5]. The nuclei showed clearing, grooving and overlapping patterns. Calcification was

noted at multiple areas. The cyst wall showed fibrosis and granulation tissue. This features were suggestive of papillary carcinoma of thyroid in the thyroglossal cyst.



[Table/Fig-5]: Histopathological picture of thyroglossal duct cyst papillary carcinoma showing papillary and follicular patterns under magnification (H&E, 200x).

DISCUSSION

The most common congenital midline neck swelling is thyroglossal duct cyst and occurs in 7% of the population. It is more common in paediatric patients due to failure of the duct to involute and further gets atrophied. Most of them are found in the infrahyoid region [1]. The thyroglossal duct is a narrow tubular structure that connects the thyroid gland to the foramen cecum. The thyroglossal duct cysts usually atrophy and disappear during the eighth and tenth weeks of gestation. However, if the duct persists, because of repeated local infection or inflammation, secretion from the epithelial lining may accumulate, triggering cyst formation.

Thyroglossal duct cysts are the most common anomaly arising from the congenital remnants of the thyroglossal duct formed during the embryogenesis of the thyroid gland. The incidence varies greatly with the age from 70% in children to 7% in adults. Only 1% of the thyroglossal duct cyst convert into carcinomas [2,3], the first description of this was given by Brentano and Uchermann in the 20th century [4]. Papillary adenocarcinoma comprises 75%-85% of the tumours reported with mixed papillary, follicular carcinomas comprises 7%, squamous cell carcinoma comprises 5%, follicular carcinoma, Hürthle cell carcinoma, and anaplastic carcinoma encountered in the decreasing order of incidence. The variant of anaplastic carcinoma leads to poor prognosis [5,6]. Various theories have been proposed for the cancerous lesion in thyroglossal duct cyst which includes a de novo origin from the rests of thyroid tissue present within the cyst [7]. The second proposed metastatic theory suggests that metastasis from the occult primaries of papillary carcinoma of the thyroid gland [8]. The only primary thyroglossal cyst tumour as well as the only true carcinoma that arises from the thyroglossal duct lining is squamous cell carcinoma [5]. Peretz A et al., suggests clinical features similar to thyroglossal duct cyst, similar USG features between thyroglossal duct cyst carcinoma and inflammatory thyroglossal duct cyst, and true positive rate of 53% by FNAC biopsy to be the possible causes for preoperative diagnosis [9]. In the presence of a normal thyroid gland, a well-differentiated carcinoma of the thyroglossal duct can be managed adequately by the Sistrunk operation. In addition to the Sistrunk operation, total thyroidectomy is preferred in advanced diseases with or without neck dissection, followed by radiotherapy and hormonal therapy. If the disease is treated adequately, the prognosis can be good.

CONCLUSION(S)

For thyroglossal duct cyst carcinoma, Sistrunk operation is sufficient in the presence of a normal thyroid gland clinically and radiographically with low recurrence. Adequate excision results in good prognosis. Because of its rare condition, the diagnosis of thyroglossal duct cyst carcinoma can be missed. Necessary investigations like radiological imaging of the neck and FNAC are required for rapidly growing midline neck swellings. Physicians need to perform a comprehensive study on the lesion including clinical and histological examinations with proper imaging to differentiate it from other midline swellings and neoplasms.

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