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Letter to Editor

Surgery Section

Thyroglossal Duct Cyst Carcinoma: A Surgical Enigma

PRIYADARSHAN ANAND JATEGAONKAR

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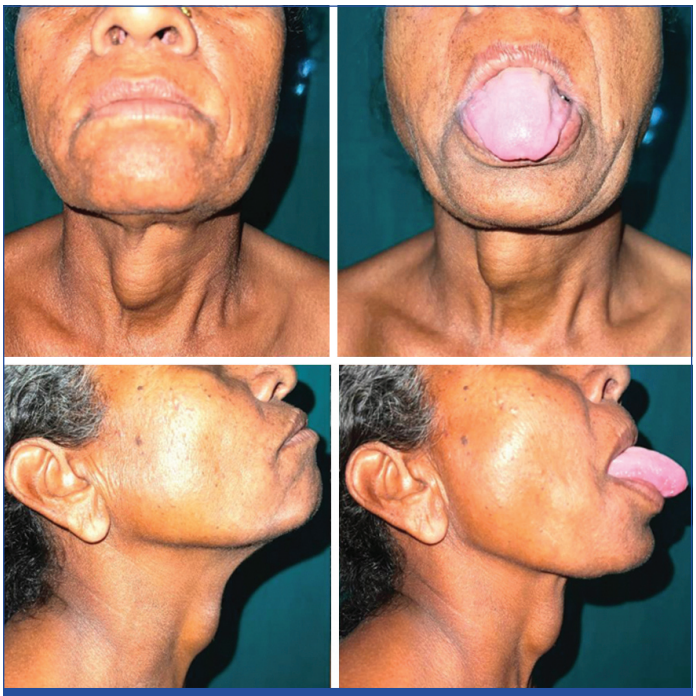
Dear Editor,

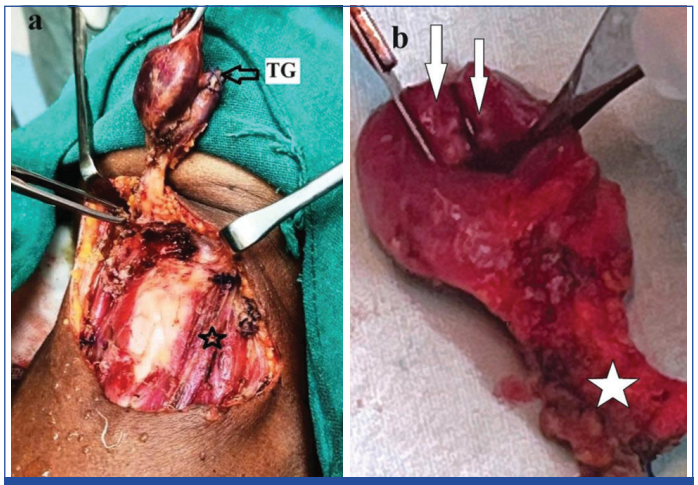
A 62-year-old woman, with no medical co-morbidities, presented with a midline neck swelling that had evolved over eight years. She denied neck irradiation in childhood and had neither associated hypo- nor hyperthyroidism nor any pressure symptoms. Examination revealed a 4×3×2 cm non tender, firm, globular mass with a smooth surface and regular margins. It moved up on deglutition and tongue protrusion. There was no cervical lymphadenopathy or thyroid nodules [Table/Fig-1].



[Table/Fig-1]: Clinical findings: Anterior and lateral neck views showing a single midline swelling with a smooth surface and well-defined margin moving up with tongue protrusion and confirming the diagnosis of Thyroglossal Duct Cyst (TGC) without any associated thyromegaly or lymphadenopathy.

Her routine haemogram and thyroid functions were normal. Neck ultrasound confirmed a subhyoid solitary well-circumscribed fluid-filled swelling without solid components or microcalcifications; the thyroid was normal without enlarged lymph nodes. Fine-Needle Aspiration Cytology (FNAC) was negative for malignant cells. Thus, with a provisional diagnosis of a simple Thyroglossal Duct Cyst (TGC), she underwent Sistrunk's operation involving en-mass excision of the cyst, the track, and the midhyoid bone. However, some areas of suspiciously thickened cyst wall were noticed during the 'back-table' dissection [Table/Fig-2]. Postoperatively, she recovered well and was discharged on the third day. Histopathology showed well-differentiated Papillary Thyroid Carcinoma (PTC) with clear margins and no lymphovascular invasion. Over the last six months, she had no locoregional recurrence.

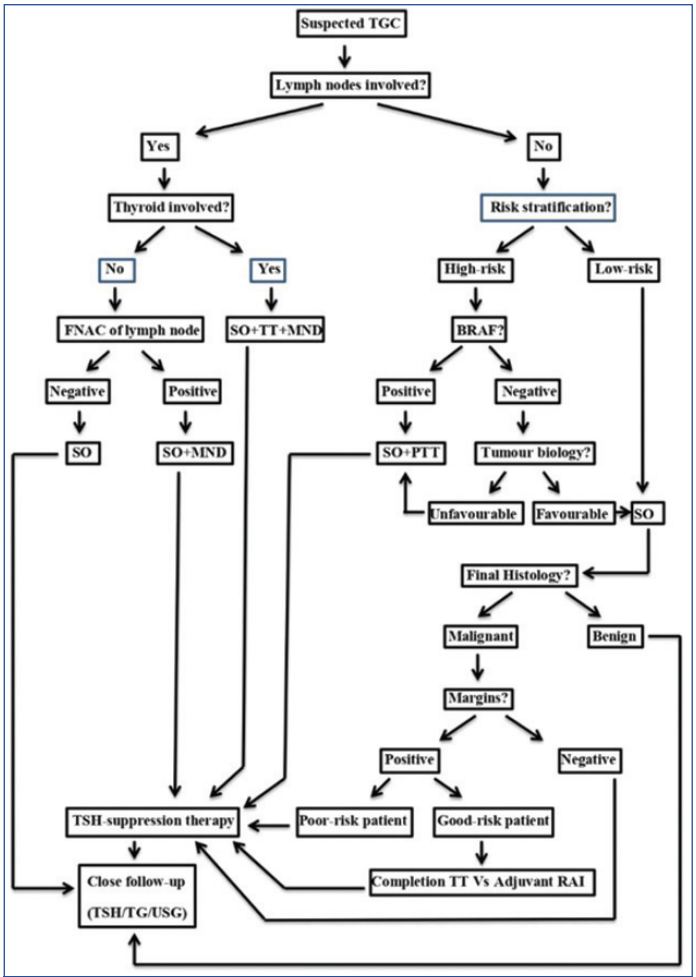
TGC carcinoma is an extremely rare condition with just 250 cases reported so far [1,2]. Over 85% of them are PTCs [1,2]. It shows a female preponderance, with a mean age of presentation at



[Table/Fig-2]: (a) Sistrunk's operation: Thyroglossal Duct Cyst (TGC) with the entire track and hyoid bone's mid-portion was excised. However, healthy strap muscles (black star) were free from any invasion and displayed no thyromegaly, and (b) Cut-open specimen: Unexpected occasional wall thickening (white arrows) with intact Thyroglossal Duct (white star).

40 years. In a recently published paper, Houas J et al., described a series of three cases, of which only one was suspected to have TGC preoperatively. However, all of them underwent Sistrunk's operation supplemented by total thyroidectomy and cervical neck dissection with good results [1]. The stark clinical resemblance makes TGC indistinguishable from benign cysts [1,2]. The two theories of its pathogenesis-de novo from ectopic thyroid tissue or metastasis from an occult thyroid malignancy-remain debatable [2]. Rapid progression, hard consistency, limited mobility, and satellite cervical lymphadenopathy are the clinical pointers of malignancy. Its irregular margins, solid components with central vascularity, and microcalcifications are reliable imaging criteria of malignancy [3,4]. However, with just 47% positive predictive value, FNAC has a limited diagnostic role, probably due to cellular dilution [3]. Like in the present case, most of the TGCs are diagnosed accidentally, only after postoperative histological examination, escalating the challenges further [2]. Positive staining of thyroglobulin, cytokeratin-19, and BRAFV600E mutation on immunohistochemistry are recently described diagnostic markers with remarkable prognostic value [2].

With no definite management consensus, most researchers agree to stratify TGC patients into low- and high-risk groups before tailoring their management [1,2,4]. Patients who are <40 years and have a cyst size <4 cm, clear margins, no pericystic infiltration, with well-differentiated biology without lymphovascular invasion or lymph node/thyroid involvement are categorised as low-risk; and vice versa are defined as high-risk [1-3]. With five-year survival approaching 100%, a meticulously performed Sistrunk's operation comprising en-mass excision of the cyst, the entire track, and the hyoid mid-body is considered adequate for low-risk persons [1,4,5]. Yet, treating high-risk patients remains contentious with plausible additions of total thyroidectomy and neck dissections [3,4]. Hence, based on the recent reports [1-5], authors propose a step-wise management algorithm for treating TGC patients [Table/Fig-3].



[Table/Fig-3]: Proposed algorithm for managing TGC carcinoma.
TGC: Thyroglossal duct cyst carcinoma; SO: Sistrunk's operation; TT: Total thyroidectomy; MND: Modified neck dissection; PTT: Prophylactic total thyroidectomy; BRAF: BRAFV600Emutation; RAI: Radio-active iodine therapy; TSH: Thyroid stimulating hormone; TG: Thyroglobulin; USG: Ultrasonography.

In conclusion, this paper aims to increase awareness about TGCs to aid informed tailored decisions with the aim of achieving improved outcomes. However, further large-volume studies are required for better understanding.

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PARTICULARS OF CONTRIBUTORS:
1. Professor, Department of Surgery (Surgical Gastroenterologist and Advance Laparoscopic Surgeon; Division of GI and Minimal Access Surgery); Pravara Institute of Medical Sciences, Loni, Rahata, Ahmednagar, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Priyadarshan Anand Jategaonkar,
403, Royal Heritage, Shrinivas Colony, Ramnagar, Wardha-442001, Maharashtra, India.
E-mail: jategaonkarpa@gmail.com

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