







Papillary Carcinoma in a Thyroglossal Cyst: A Case Report

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Abstract

Keywords

- ▶ thyroid neoplasm
- ▶ thyroid carcinoma
- ▶ thyroglossal duct cyst

We herein report a case of papillary thyroid carcinoma with an unusual presentation, inside a thyroglossal cyst. An 83-year-old male patient reported a long-standing nodule persisting for over 40 years, with progressive enlargement and periodic dysphonia. The patient underwent total thyroidectomy combined with the Sistrunk procedure. Resection of the thyroglossal duct cyst and total thyroidectomy are the treatments of choice for this rare presentation. The case herein reported is deemed atypical according to the current literature, and it was managed following the existing guidelines, resulting in a satisfactory outcome.

Introduction

The thyroid originates by the third week of gestation, and it is formed by an invagination of endodermal epithelial tissue in the anterior wall of the pharynx, migrating caudally through the midline of the neck, reaching the cricoid cartilage and the first tracheal rings around the seventh gestational week. The duct formed by the path of the thyroid from the base of the tongue to its final location gives rise to the thyroglossal duct, which generally undergoes a process of atrophy and becomes obliterated during the tenth week.¹ Failure of this mechanism results in residual epithelial tissue, which, through the production and secretion of mucus, forms the thyroglossal duct cyst (TDC).

Primary malignancies of TDC are rare, reported in less than 1% of the cases.² Papillary carcinoma is the most common histological type, accounting for 75 to 80% of the tumors reported. Other variants include squamous cell carcinoma (5%) and follicular carcinoma (1.7%).³ The etiology of TDC carcinomas is controversial and remains a subject of debate within the medical community. Two theories may explain their origin: the first is the *de novo theory*, in which

malignant cells arise from ectopic thyroid tissue in the walls of the thyroglossal duct; the second, the *metastatic theory*, suggests that a hidden primary thyroid lesion metastasizes to the thyroglossal duct.⁴

The diagnosis of TDC is established via ultrasound, which enables the visualization of the ectopic thyroid tissue. Malignancy can only be confirmed through a histopathological examination, which enables the distinction between primary and metastatic TDC carcinoma.⁵ The treatment of choice is the Sistrunk procedure, which involves resection of the entire TDC tract, including the foramen cecum and the central portion of the hyoid bone.⁶

The present study was submitted to and approved by the institutional Ethics Committee, and informed consent was obtained from all participants.

Case Report

An 83-year-old male patient, with a history of chronic alcohol use and former tobacco consumption, presented with a long-standing cystic lesion in the thyroid region, persisting for more than 4 decades and exhibiting

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progressive volumetric increase, which prompted a medical evaluation. Upon physical examination, a 2.5 cm nodular lesion was observed, freely mobile on palpation, exhibiting a bosselated contour, with no evidence of cervical lymphadenopathy. An oroscopic examination and video nasopharyngolaryngoscopy revealed no pathological findings.

Computed tomography (CT) of the head and neck showed a lesion in the right paramedian region, around the level of the hyoid bone and thyroid cartilage, with a cystic component measuring approximately 1.9 cm in the right paramedian line, along with a solid component containing internal calcifications measuring 1.7 cm in diameter, without evidence of cartilage invasion. Ultrasonography (USG) of the thyroid gland showed an enlarged gland with normal echotexture and mixed, hypoechoic, well-defined nodules without microcalcifications. Two nodules located in the left thyroid lobe demonstrated a benign appearance (of category 2 according to the Thyroid Imaging Reporting and Data System, TIRADS), measuring approximately $1.1 \times 1.0 \times 0.7$ cm and $0.6 \times 0.6 \times 0.3$ cm. A solid-cystic cervical nodule within the thyroglossal duct cyst, previously identified on CT, was also visible. (►Figs. 1, 2)

Fine-needle aspiration cytology (FNAC) was performed on a tumor lesion located in the right paramedian region of the neck, and it revealed findings consistent with ectopic papillary thyroid carcinoma. (►Fig. 3)

A core-needle biopsy guided by cervical ultrasound was performed on the nodule located in the right paramedian midline, presenting an image compatible with a thyroglossal duct cyst. A histopathological examination revealed malignant characteristics, consistent with infiltrative papillary

thyroid carcinoma. As treatment, the Sistrunk procedure and total thyroidectomy were performed. (►Fig. 4)

The histopathological examination revealed a TDC containing a papillary thyroid carcinoma measuring 2.7 cm, infiltrative, surrounded by a fibrous capsule, with mild nuclear atypia and capsular invasion. The tumor was well-differentiated (grade 1/4) (►Fig. 5).

In the left thyroid lobe, a thyroid neoplasm along with uninodular hyperplasia were pinpointed, measuring less than 1 cm. These findings suggest noninvasive follicular thyroid neoplasm with papillary-like nuclear features (NIFTP). The neoplasm displays nuclear traits suggestive of papillary thyroid carcinoma or may represent metastatic dissemination of papillary carcinoma from a TDC.

The histopathological evaluation was inconclusive in differentiating between these diagnostic possibilities. No significant pathological alterations were observed in the isthmus or right thyroid lobe.

Ninety days after thyroidectomy, and prior to radioiodine therapy administration, the patient's serum thyroglobulin concentration was <0.10 ng/mL, the level of anti-thyroglobulin antibody was of 11.9 IU/mL, and the concentration of thyroid-stimulating hormone (TSH), of 0.78μ -IU/mL. Then, the patient underwent radioiodine ablation therapy with a therapeutic dose of 100 mCi of iodine-131. Whole-body scintigraphy revealed a discrete focal area of radiotracer uptake, consistent with residual thyroid tissue (►Fig. 6).

Discussion

The first case of this form of carcinoma was reported by Brentano in 1911 and by Hassan et al.² in 2016, involving the lingual thyroid. Since then, only a restricted number of

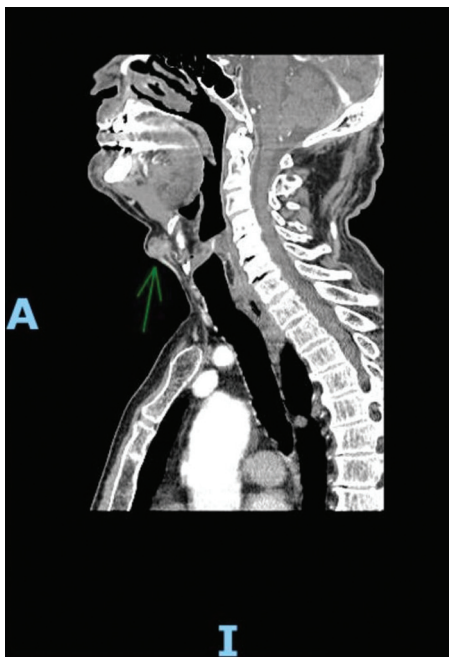


Fig. 1 Sagittal computed tomography scan showing the thyroglossal cyst, a solid-cystic tumor lesion of the anterior cervical soft tissues of an altered malignant neoplastic nature.

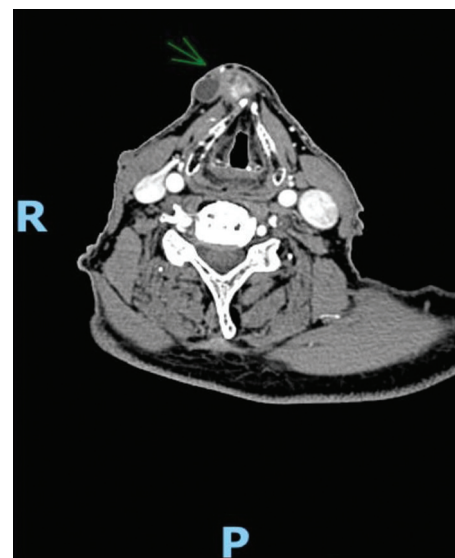


Fig. 2 Axial computed tomography scan showing the thyroid gland with a right paramedian cystic component measuring 1.9 cm and a solid component with enhancement and internal calcifications measuring 1.7×1.7 cm, with fascia infiltration.

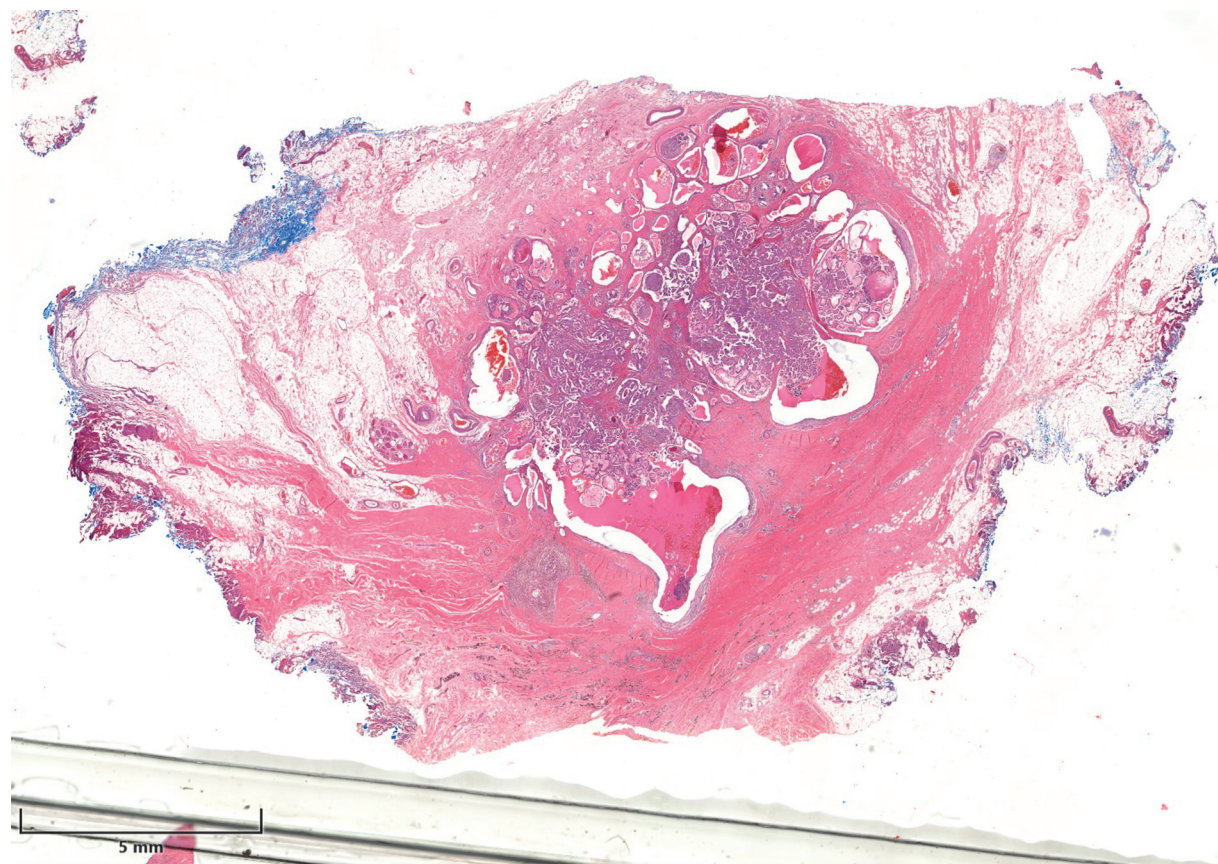


Fig. 3 Hematoxylin and eosin-stained section – A centrally located papillary neoplasm is identified, characterized by irregular contours. The neoplastic proliferation is surrounded by dense fibrotic stroma. Papillary structures will be more distinctly visualized in subsequent high-magnification images.

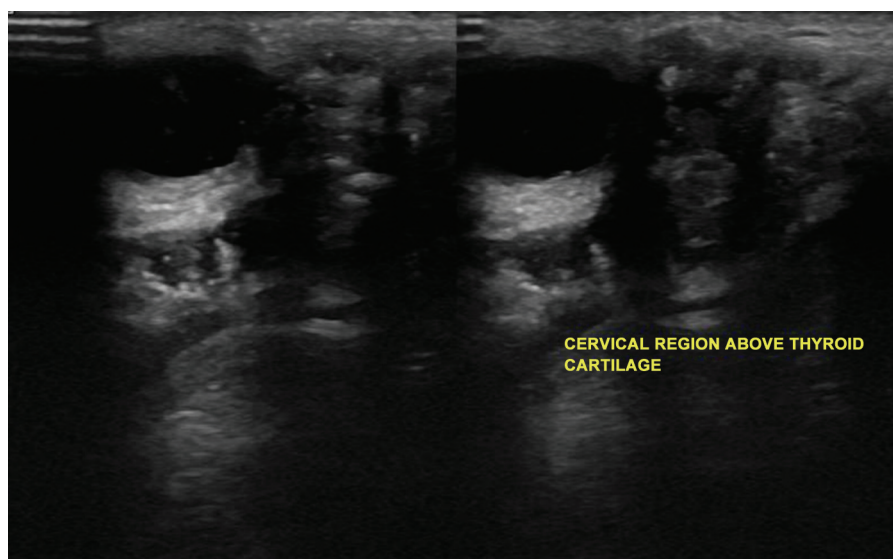


Fig. 4 Neck ultrasound scan, with a solid-cystic nodule located in the midline, inferior to the hyoid bone, presenting internal calcifications and measuring approximately 3.6 cm.

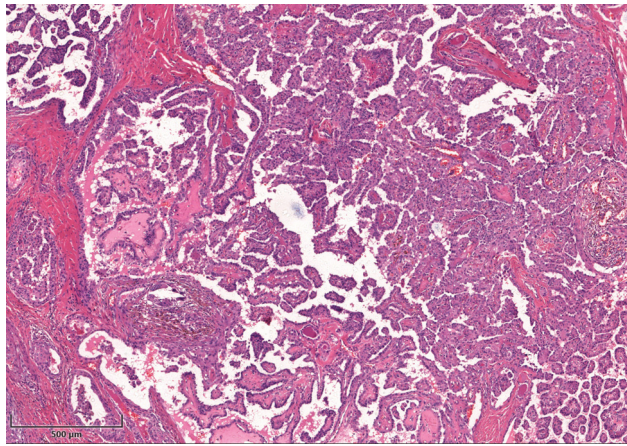


Fig. 5 Hematoxylin and eosin-stained section – Intermediate magnification of the central region highlighting the papillary architecture. At this magnification, the papillary structures are distinctly visualized and readily identifiable.

these cases have been described in the literature, since papillary carcinoma arises from a thyroglossal duct cyst and is deemed a rare pathological entity, with fewer than 300 cases documented worldwide.² According to the current literature, the prevalent histological subtypes of TDC carcinomas are: papillary carcinoma (80%), mixed papillary and follicular carcinoma (8%), and squamous cell carcinoma (6%).²

Thyroglossal duct cysts should be investigated upon the recognition of an inflexible, immobile, irregular mass. Computed tomography, fine-needle aspiration, magnetic resonance imaging and USG are commonly employed during the investigative process.⁶

The management of this carcinoma remains a subject of debate within the medical community. The Sistrunk procedure is considered by most authors to be the optimal treatment modality; however, some experts recommend the inclusion of total thyroidectomy, given the high incidence of concomitant thyroid neoplasms.⁷ Total thyroidectomy, regardless of the evidence of thyroid involvement, is further supported by other studies.

The reasoning for thyroid resection in patients with TDC carcinoma is based on three factors: the possibility of malignancy within the thyroid gland; the need for radioiodine therapy; and the use of serum thyroglobulin levels as a biomarker for thyroid carcinoma.¹ Based on these three factors, the patient herein described underwent total thyroidectomy.

Regional lymph node metastases have only been reported in 7.7% of the cases.⁸ Distant metastases are rare, occurring in less than 2% of the cases. This generally leads to a favorable prognosis when appropriate management is implemented.³ For the posttreatment surveillance of TDC, patients are advised to undergo annual follow-up, including cervical USG and an assessment of the level of serum thyroglobulin.¹

It is also necessary, if possible, to differentiate primary and metastatic lesions in TDC carcinoma. Primary TDC carcinomas are typically defined by three characteristics: the presence of carcinoma within the cyst wall; the identification of normal thyroid tissue adjacent to the tumor; and the absence of primary carcinoma in a clinically- and radiologically-normal thyroid gland.⁵

In the case herein reported, the thyroid gland presented clinical and radiological alterations, leading to the decision to perform a Sistrunk procedure in combination with total thyroidectomy and radioiodine therapy. Radioiodine therapy is particularly indicated in cases of suspected or confirmed metastatic disease.⁵

The clinical implications of this disease suggest that most cases of papillary carcinoma of s TDC are diagnosed incidentally, postoperatively, as benign and malignant lesions are often clinically indistinguishable.⁹ Therefore, it is essential to rule out secondary lesions or metastatic disease, which is crucial for appropriate treatment and better clinical outcome.⁵ The optimal management requires a multidisciplinary approach, involving surgery, endocrinology, nuclear medicine, and clinical follow-up.¹⁰

In conclusion, the current case report contributes to the limited literature on papillary carcinoma arising from a TDC, reinforcing the importance of continued research, detailed case reporting, accurate diagnosis, and the necessity for the

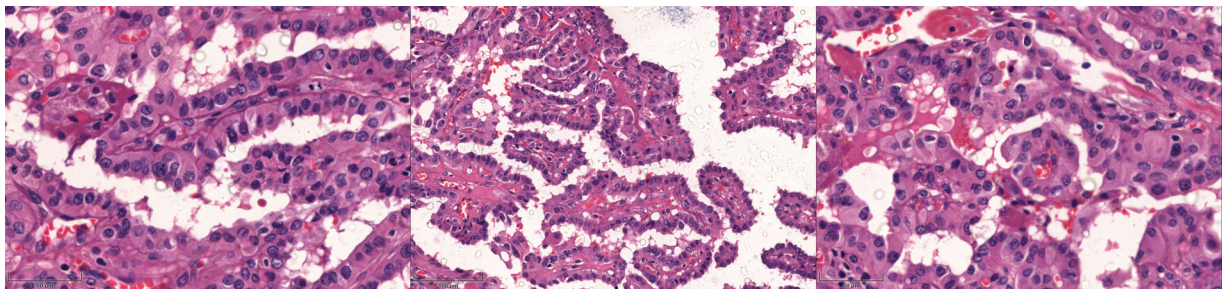


Fig. 6 Hematoxylin and eosin-stained section – High magnification of the papillary structures. The papillae are lined by cells exhibiting mild atypia and slight anisokaryosis. Occasional nuclear grooves and nuclei with central chromatin clearing are also observed.

development of more comprehensive treatment strategies for this pathology.

Authors' Contribution

BHN: collection and assembly of data, data analysis and interpretation, manuscript writing; GGP: collection and assembly of data, manuscript writing; LGLS: collection and assembly of data, manuscript writing; GMN: collection and assembly of data, data analysis and interpretation, manuscript writing, conception and design, final approval of the manuscript; AA, data analysis and interpretation, manuscript writing, conception and design, final approval of the manuscript; HMN: data analysis and interpretation, manuscript writing, conception and design, final approval of the manuscript, provision of study materials or patient involvement.

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Conflict of Interests

The authors have no conflict of interests to declare.

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