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Case report

Tall cell carcinoma arising in a thyroglossal duct cyst: A case report

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HIGHLIGHTS

- TDCCs are uncommon but usually with a good prognosis.
- An association with thyroid primitive carcinoma is frequently observed.
- An extremely rare TDCC variant with a poor prognosis associated with two synchronous foci of thyroid carcinoma is reported.
- Surgical management of the thyroid gland within treatment plan is controversial.
- An accurate preoperative evaluation is mandatory for an optimal management.

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ABSTRACT

Introduction: Thyroglossal duct cyst carcinomas are extremely rare and their clinical presentation is similar to that of benign cysts. The diagnosis is based on physical examination, laboratory tests, and most importantly multiple imaging techniques (ultrasonography, computed tomography and magnetic resonance imaging), and fine needle aspiration cytology.
Presentation of case: We report a very unusual case of a tall cell variant of papillary carcinoma arising in a thyroglossal duct cyst in association with a follicular variant of papillary microcarcinoma and a tall cell variant of papillary carcinoma arising from the thyroid gland.
Discussion: Although rarely described in the medical literature, ectopic thyroid tissue present in the thyroglossal duct cyst could be involved in the development of a poorly differentiated carcinoma. The frequent observation of an associated primitive thyroid carcinoma makes surgical management of thyroid gland controversial.
Conclusion: For the optimal management of this rare pathological condition, a comprehensive preoperative evaluation and meticulous intra-operative appraisal are fundamental.

1. Introduction

The thyroid gland originates from a median endodermal thickening in the floor of the primordial pharynx at the base of the tongue. This thickening soon becomes a small outpouching that descends into the neck during its maturation. This thyroid primordium is connected to the tongue by the thyroglossal duct a narrow epithelial tube that degenerates and disappears before the definitive development of the thyroid gland. Various thyroid pathologies are known both in thyroid gland and in the ectopic thyroid tissue [1].

The thyroglossal duct cyst is a cystic expansion occurring due to an abnormality in thyroid development resulting from the defective obliteration of the duct during embryogenesis. A persistent thyroglossal duct is usually a benign condition, but in some cases it may undergo dilatation and be responsible for cyst formation, which clinically presents as a mass arising along the thyroglossal tract [2]. This abnormality occurs in around 7% of the population [3] and it represents the most common congenital anomaly of the neck resulting in more than 75% of median neck swellings in childhood. Between 1.5 and 45% of cases show the presence of ectopic thyroid tissue [2,4].

The development of a carcinoma of the thyroglossal duct cysts is rather rare, occurring in approx. 1% of cases. The median age at

Abbreviations: TDCC, thyroglossal duct cyst carcinoma; US, ultrasonography; CT, computed tomography; MRI, magnetic resonance imaging.

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presentation is 40 years and females are affected more often than men with a 3:2 ratio [5,6]. The peak incidence is in the third decade for women and sixth decade for men and it presents an overall age and sex distribution similar to that of thyroid carcinoma. Regional lymph node metastases are less common in the thyroglossal duct cyst papillary carcinoma compared to a primary cancer of the thyroid, with an incidence <8% [2]. Furthermore, papillary duct cyst carcinoma rarely presents distant metastases and its prognosis is similar to that of the thyroid papillary carcinoma.

The vast majority of thyroid cancers concern papillary carcinomas (79.9%), followed by much fewer cases of follicular or mixed follicular-papillary (9.5%), squamous cell carcinomas (7.6%), anaplastic carcinomas (0.6%), and Hürthle cell carcinomas (0.6%) [7]. No reports of medullary carcinoma exist to date, probably due to the fact that C cells originate from the last pharyngeal pouch, supporting the theory of the primitive genesis of carcinomas arising in ectopic thyroid tissue rather than from the metastatic diffusion of thyroid carcinoma through the thyroglossal duct [8]. On the other hand, a synchronous papillary carcinoma on the thyroglossal duct cyst and on the thyroid gland can be explained as a multi-focus manifestation of the tumor and not as metastatic foci [9]. An association with primitive thyroid papillary carcinoma is observed in about 25–40% of patients undergoing total thyroidectomy as part of their surgical treatment [6,9].

Papillary carcinoma is therefore the most frequent malignancy arising from ectopic thyroid tissue of this remnant; moreover, a tall cell variant is extremely rare and shows a rather poor prognosis. Compared with classical papillary thyroid carcinoma, tall cell variant had a higher rate of extrathyroidal extension (53.6% vs. 30.2%), and a poorer 5-year disease-specific survival (81.9% vs. 97.8%) [10]. Differential diagnosis includes Hürthle cell variant, oncocytic papillary carcinoma and Warthin-like papillary carcinoma [9,11].

To date, only one case of tall cell carcinoma arising in a thyroglossal duct cyst has been reported in the medical literature [11]. Here, we present a remarkable case of tall cell carcinoma arising in a thyroglossal duct cyst associated with a follicular variant of papillary microcarcinoma and a tall cell variant of papillary carcinoma of the thyroid gland.

2. Presentation of case

A 77-year-old Caucasian woman was admitted to our Department with recent onset (2 months) of a median neck swelling.

She did not have any obstructive symptoms, such as dysphagia, dyspnoea, stridor or dysphonia.

The patient had a history of essential hypertension, dyslipidemia and type 2 diabetes mellitus. She had also been hospitalized in the past for a transient ischemic attack.

Physical examination revealed a voluminous swelling of about 8 cm, extending from the hyoid region to the jugular notch and laterally within the anterior margin of the sternocleidomastoid muscle. The swelling was oval in shape and with a smooth surface; it was mobile, firm and painless (Fig. 1).

Thyroid functional tests (serum thyroxine, triiodothyronine and thyroid stimulating hormone), calcitonin and thyroid antibodies were within normal limits.

A preoperative ultrasound neck evaluation showed a voluminous anterior midline neck mass of about 7–8 cm with an inhomogeneous echostructure and an isoechoic solid component (largest diameter of about 3–4 cm); high intranodular flow by Doppler analysis was associated with an hypoechoic solid nodule at the level of the thyroid isthmus of the thyroid gland with microcalcifications inside. No regional suspect nodes were identified.

The results of fine needle aspiration cytology suggested possible malignancy.

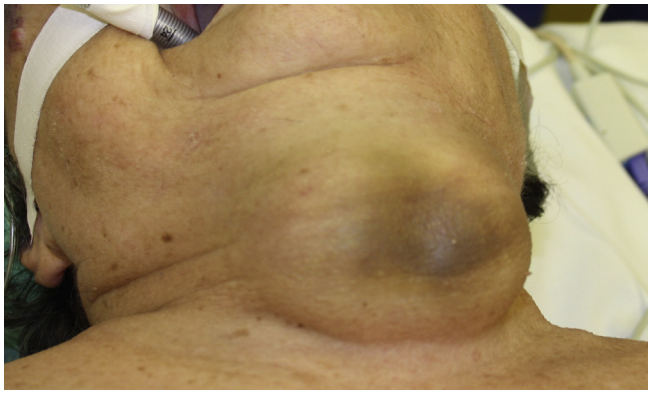

Fig. 1. Preoperative image: the large cervical swelling is visible.

Intralesional thyroglobulin levels were 27.550 ng/ml (normal values: <10 ng/ml).

The patient underwent mass and thyroglossal duct excision and total thyroidectomy. Parathyroid glands and recurrent laryngeal nerves were identified and preserved.

The surgical specimen included a skin lozenge with midline neck mass and the thyroid gland (Fig. 2).

Microscopic examination revealed the presence of an infiltrative tall cell variant of papillary carcinoma, arising in the thyroglossal duct cyst in conjuction with a follicular variant of papillary microcarcinoma of the left thyroid lobe and a peri-isthmic tall cell variant of papillary carcinoma (Figs. 3–5). No evidence of cervical lymph node metastases was found.

The patient then underwent radioiodine ablation therapy (106 mCi).

Thyroglobulin level after radioiodine ablative therapy was <0.20 ng/ml (normal values: 0.73–84.00 ng/ml).

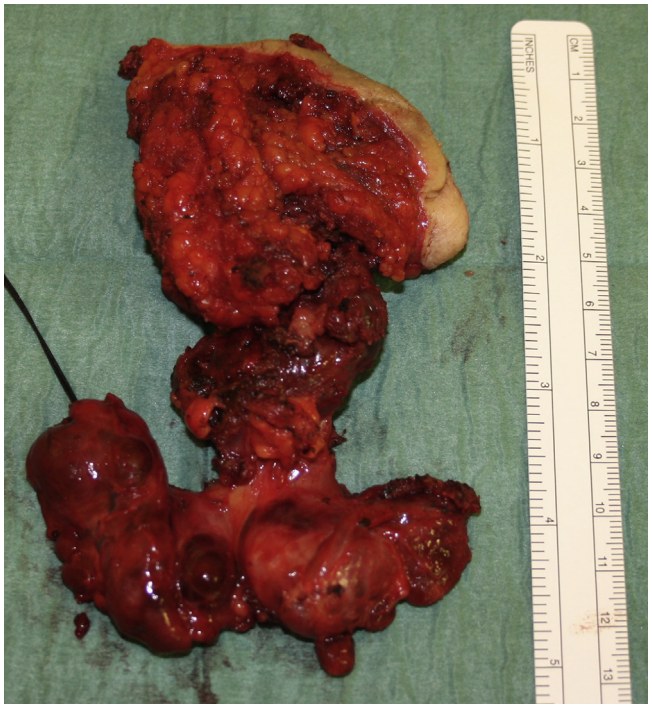

Fig. 2. Surgical specimen: thyroid, neoplasm, thyroglossal duct along with a lozenge of skin.