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New Case of Papillary Thyroglossal Tract Cyst Carcinoma, in the Absence of Clear Guidelines, What Should Be Do?

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Abstract **Case Report**

Thyroglossal tract cyst carcinoma (TTCC) is a rare pathological situation, The challenge is mainly in the surgical management. In this article, we report a new case of papillary TTCC in a 51 years old female patient, not suspected preoperatively and confirmed postoperatively by histological analysis of the resected specimen. The therapeutic strategy was completed by a total thyroidectomy with adjuvant treatment with radioactive iodine and hormone restraint therapy. The outcome was favorable after 7 years of follow-up. In the absence of clear guidelines, the management of TTCC depends on the clinical situation and the experience of the treating team.

Keywords: Papillary carcinoma, thyroglossal tract cyst, thyroid surgery, Ira therapy.

INTRODUCTION

TTC are congenital cervical malformations, the occurrence of papillary TTCC is rare even if its incidence is not exact. The diagnosis is only made postoperatively and its management is not well codified.

OBSERVATION

A 51 years old female patient with no particular pathological history, consulted for an anterior cervical swelling progressively increasing in size over the past 8 years and becoming compressive. Clinical examination revealed a patient in good general condition with a median cervical mass, firm in consistency, painless, measuring 3x3 cm, rising on swallowing without inflammatory signs or cervical adenopathy. Ultrasound showed an anterior cervical mass measuring 28x22 mm in diameter, with a heterogeneous echo structure containing hyperechoic debris, raising the suspicion of a TTCC. The thyroid gland was normal with no detectable cervical adenopathy. The patient was operated on using the Sistrunk technique. Histopathological examination of the surgical specimen revealed a papillary carcinoma on a TTCC (Figure 1,2). A total thyroidectomy and lymph node dissection were therefore performed, but the histopathological study did not reveal any thyroid neoplasia. A whole-body isotope scan with iodine 131 did not reveal any thyroid tumor residues or distant metastases. Ira therapy was prescribed at a dose of 100 mCi, with hormone therapy at a restraining dose. The evolution was favorable: no recurrence and no metastases after a 6-year follow-up.

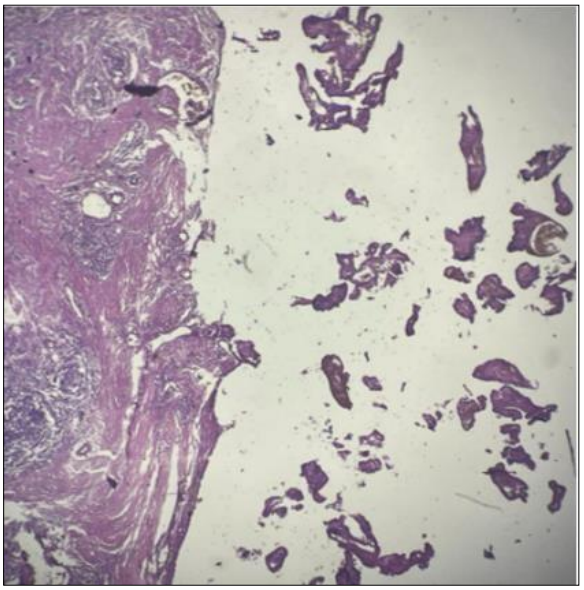

Figure 1: Morphological appearance showing a papillary tumor proliferation at the edge of the cystic wall.

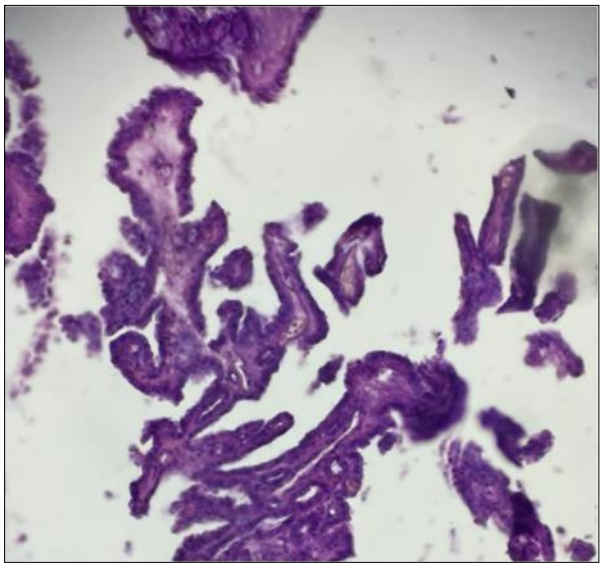

Figure 2: Morphological appearance in Gx20 showing papillae lined by cells with non-incised, overlapping and clarified nuclei characteristic of papillary thyroid carcinoma.

DISCUSSION

TTCs are due to abnormal persistence of the thyroglossal duct; an embryonic remnant of the migration of the thyroidal precursor from the base of the tongue to its final anatomical location [1]. Autopsy studies have shown that they have up to 62% ectopic thyroid tissue, which can then degenerate.

The incidence of TTCC is unknown, probably in the order of 1%. Since Brentano's first description in 1911, only about 300 cases have been published, mostly as case reports or small series [2].

The papillary type is the most common and is found in about 83% of cases. The other types are mixed papillary-vesicular carcinomas (8%), squamous cell carcinomas (6%), a few cases of Hûrthle cell, vesicular, anaplastic and squamous cell carcinomas. No cases of medullary carcinoma have been described [3].

The tumorigenesis of TTCC is not well understood, two hypotheses are proposed: the first is that carcinomas develop de novo on the TTCC due to the presence of thyroid tissue at the level of the cyst [4], the second is that the thyroglossal duct is a natural route for the spread of thyroid neoplasia [5].

The clinical presentation of TTCC is not specific. Most patients present with a painless mobile medial or paramedian swelling, which cannot be distinguished from a normal medial cervical cyst [6].

Magnetic resonance imaging is rarely used in the preoperative evaluation of suspected TTCC. However, if it is performed, the presentation is an invasive peripheral solid mass within the cyst or along the thyroglossal tract with or without adenopathy [7].

Due to the rarity of this tumor, no standardized procedure could be established, especially on the need for total thyroidectomy, lymph node dissection and radioactive iodine treatment [2].

In all cases, excision of the cyst should be performed according to the Sistrunk technique (from the medial part of the body of the hyoid bone to the nucleus of the tissue surrounding the thyroglossal tract at the foramen caecum) [8].

Total thyroidectomy is still controversial [2], Pfeiffer *et al*., consider that the Sistrunk technique associated with lymph node dissection are sufficient for patients who have no thyroid signs of malignancy on imaging, they consider that total thyroidectomy does not significantly improve survival and exposes the patient to postoperative risks and lifelong hormonal treatment [2]. Other teams advocate total thyroidectomy for a number of reasons: the possibility of coexisting thyroid neoplasia [9], the possible use of ablative Ira therapy and the ease of biological (Thyroglobulin assay) and isotopic follow-up [10].

There is also debate about lymph node dissection, with some teams only performing it when there are suspicious adenopathies on imaging [11, 12], while others do it systematically as a prophylaxis [13].

Papillary TTCC should be considered as distinct and highly differentiated tumors. Therefore, it seems logical to perform a recurrence risk stratification comparable to that used for well- differentiated thyroid carcinomas [14]. The criteria useddiffer slightly, but are broadly consistent with the American Thyroid Association parameters for differentiated thyroid carcinoma. Patients under 45 years of age, with small tumors (cut-off values here vary from1 to 4 cm) without peri-cystic soft tissue infiltration or angio-invasiveness, healthy resection margins and no cervical or thyroid abnormalities on imaging, are considered low-risk groups. All other patients are classified in the high-risk group and undergo more aggressive treatment: total thyroidectomy + lymph nodedissection + radioiodine.

Braking treatment is indicated in all patients [3]. Follow-up is the same as for differentiated thyroid carcinomas [2-15].

The prognosis of TTCC is very good with a low risk of locoregional recurrence. Only a few isolated cases of distant metastasis or death associated with the disease have been reported [2]. Recurrences may occur years or even decades later, so effective lifelong surveillance is required [5].

CONCLUSION

TTCC is a rare entity with an overall good prognosis. Its management is still a matter of debate between those who are satisfied with excision of the cyst and others who opt for a more aggressive treatment. In all cases, it is necessary to stratify the risk of recurrence in order to identify the modalities of subsequent follow-up. With this work, we add a new observation of a rare carcinoma with the expectation that further studies will be done to standardize the therapeutic procedure and improve the prognosis.

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