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Rare Localization of Papillary Thyroid Carcinoma on Thyroglossal Tract Cyst

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Abstract **Case Report**

Thyroglossal tract cysts (TTCs) are common and degeneration is rare, occurring in only around 1 to 1.5 % of cases. Papillary carcinoma is the most common histological type. The clinical presentation of these cancers is very often non-specific, and their diagnosis is usually made postoperatively. Controversy surrounds the management of this type of neoplasia. We report the case of a 34-year-old patient who underwent surgery for an anterior cervical swelling that had been present since childhood, and whose histological examination was consistent with papillary carcinoma on KTT. He also underwent total thyroidectomy and iratherapy, and was put on a hormone suppression treatment even though the thyroid was unaffected, in the absence of consensus concerning this pathology. Progression after one year's follow-up remained favourable.

Keywords: Thyroglossal tract, cyst, papillary, carcinoma.

INTRODUCTION

Thyroglossal tract cyst (TTC) is a rare condition associated with congenital cervical malformation, due to abnormal persistence of the thyroglossal duct [1, 2]. Degeneration is rare, occurring in 1 to 1.5% of cases [1, 3, 4], and is most often papillary carcinoma. The clinical presentation is non-specific, and the finding of cancer at this level is always a histological discovery. Management remains controversial. We report the case of a patient operated for papillary carcinoma on a cyst of the thyroglossal tract.

CASE REPORT

This was a 34-year-old patient with no previous pathological history, who consulted us for an anterior cervical swelling that had been progressively increasing in size since childhood, with no signs of compression, but was becoming cosmetically bothersome. Clinical examination revealed a patient in good general condition, with a firm, painless, median anterior cervical mass measuring 3cm by 2cm, rising on swallowing and tongue protraction, with no inflammatory signs or cervical adenopathies (figure 1). A cervical CT scan revealed a 34*19*27mm multi-loculated median anterior subhyoid cervical formation with cystic and fleshy components, with microcalcifications most probably related to a suspected complicated thyroglossal tract cyst (figure 2). The patient underwent complete excision, removing the cyst and the body of the hyoid bone. Histopathological examination of the surgical specimen revealed a papillary tumour proliferation with a morphological appearance suggestive of papillary carcinoma on remodelled TTC without neoplastic vascular emboli. A follow-up cervical ultrasound revealed a largely cystic left lobar thyroid micronodule graded Eu-Tirads 2 with a small left paramedian adenomegaly measuring 5.3*4.6*3.3mm, above the operative scar at a distance of 15 mm, suspicious in appearance and not accessible to cytopunction. After a multidisciplinary discussion, the patient underwent total thyroidectomy with prelaryngeal central curage; the anatomopathological study revealed a morphological appearance in favor of a diffuse dystrophic goiterremodeled with striated conjunctivo-muscular tissue without histological signs of malignancy. Scintigraphy with iodine131 revealed 2 cervical fixation foci suggestive of thyroid residues. The patient was referred to nuclear medicine for further IRA therapy. He was put on L-Thyroxine at a suppressive dose, and remission was declared after 2 courses of 100 mCi iodine131.

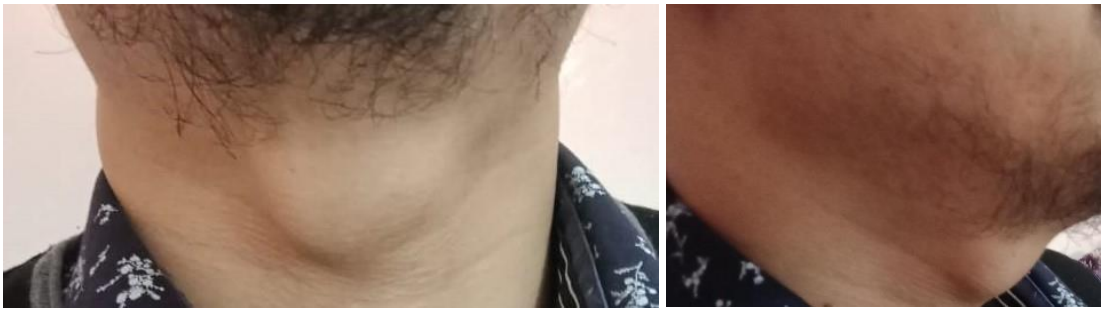

Figure 1: Subhyoid TTC in our 34-year-old patient

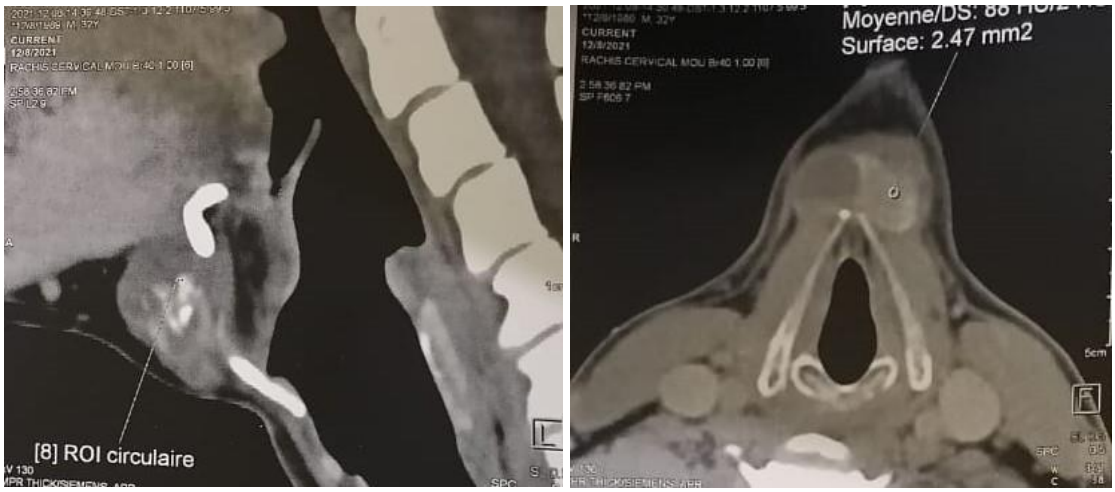

Figure 2: A subhyoid mass measuring 34*19*27mm, with cystic and fleshy components, site of microcalcifications related to a suspected complicated thyroglossal tract cyst

DISCUSSION

The thyroglossal tract cyst (TTC) is a congenital cervical malformation, due to abnormal persistence of the thyroglossal duct, an embryonic remnant of the migration of the thyroid roughening from the base of the tongue to its definitive anatomical location [1, 5].

This TCC may be located along the entire tract, from the base of the tongue to Lalouette's pyramid [6]. Congenital cervical malformations are common in paediatric ENT, accounting for 40% of all congenital cervical malformations. They may persist into adulthood [6].

The risk of malignant degeneration from thyroglossal tract cysts remains rare. It occurs in 1 to 1.5% of cases. But it is certainly underestimated, as preventive excision surgery is very often performed in childhood [7].

They mainly occur in adults around the fourth decade of life [1, 3, 8]. The first description of carcinoma on TTC dates back to 1911 with BRENTANO [3]. Of the cases of carcinoma on TTC reported in the literature [7], 83% are of the papillary type. Other types include mixed papillofollicular carcinomas in 8% of cases, squamous cell carcinomas in 6%, with a few cases of Hûrthle cell carcinomas and follicular, anaplastic and epidermoid carcinomas. No cases of medullary carcinoma have been described in the literature [1, 3, 8, 9]. Indeed, the distribution of the different histological types appears to be comparable to the percentage found in thyroid cancers.

Many authors believe that these carcinomas develop de novo within the TTC [10], their origin being normal thyroid tissue present both in the cyst wall and all along the path of the tract, and whose frequency varies from 1.5 to 62% of cysts [9, 11]. Other authors believe that the thyroglossal duct constitutes a natural pathway for the spread of carcinomas from the thyroid gland [12].

Some hypotheses concerning the pathophysiology of these histological types have been reported. The majority of authors believe that these lesions develop de novo within the cyst and are, therefore, primitives of the thyroglossal tract [10], their origin being normal thyroid tissue present both in the cyst wall and all along the path of the tract, and whose frequency varies from 1.5 to 62% of cysts [11, 12], while others such as Belnouesuggest that this thyroglossal tract may constitute a natural pathway for the spread of a carcinoma originally developed in the thyroid gland [12]. Which explains the different attitudes concerning the therapeutic management of this pathology [13].

The clinical presentation is generally similar to that of a simple TTC, which explains why it is most often

discovered by chance following anatomopathological examination of the surgical specimen [1,4], as in the case of our patient. However, a few clinical signs should prompt the practitioner to suspect a possible neoplastic process, in particular the hard, fixed and/or irregular nature of the cervical mass, which may have increased in size progressively, or be associated with cervical adenopathy [3].

There is controversy as to how to deal with the discovery of cancer on anatomopathological examination of the TTC excision specimen, particularly as regards the need for thyroidectomy associated with excision of the TTC [8].

According to several authors, it is recommended to complete the initial surgical procedure with a total thyroidectomy. The reasons given are the frequency of association of degenerated TTCs with primary thyroid carcinomas, which varies from 11 to 40%, and the guarantee of better follow-up, since in a metanalysis, PATEL showed that only the extent of the initial surgical procedure was a significant variable in terms of survival [1-3, 5, 8]. Thyroidectomy would be more essential in the event of tumour invasion of the cyst wall, in the event of individualization of a vesicular or epidermoid histological type or clinical or ultrasound revelation of a nodular thyroid lesion, and for some authors when there is doubt about the patient's ability to adhere to regular medical follow-up [1]. Lymph node dissection would be carried out when adenopathies are clinically or ultrasonographically suspicious [1-3]. A whole-body 131-iodine scan and thyroglobulin assay should be performed after thyroidectomy. If tumour residues are present on the scan, ablative dose iratherapy should be performed. Thyroid hormone therapy at a restraining dose and annual thyroglobulin monitoring are always indicated [1, 2]. Recurrences can occur years or even decades later, and are often demanding, requiring effective lifelong monitoring [11]. Others, such as GEOK, see no point in thyroidectomy if the thyroid is intact [12]. Their arguments are the increase in morbidity due to iterative operations, the possibility of effective follow-up with reintervention at a later stage if thyroid cancer is discovered, and finally the good prognosis of these cancers.

The prognosis of carcinomas on TTC appears to be better than that of the thyroid, due to the rarity of distant metastases [1, 3]. Survival at 5 and 10 years is estimated respectively at 100 and 95.6% according to PATEL for 62 patients with differentiated thyroid cancer on TTC; no cases of local recurrence have been described and only two patients developed lymph node metastases [3, 12]. However, long-term postoperative monitoring is mandatory.

In our patient, we opted for excision of the TTC combined with total thyroidectomy and 2 courses of iratherapy, given the persistence of 2 hyperfixing thyroid residues on iodine-131 scintigraphy and the fear of ignoring a carcinoma at the level of this residue. Treatment with L-Thyroxine was prescribed, with the aim of achieving a TSHus between 0.5 and 2 µIU/ml. Progression after one year's follow-up remained favourable.

CONCLUSION

Papillary carcinoma on TTC is a rare entity. The absence of clinical or para-clinical features to clinically differentiate an ordinary TTC from a papillary carcinoma developed on thyroid remnants necessitates primary surgical treatment with histological study of all TTCs. The problem lies in the subsequent therapeutic management and follow-up in the absence of consensus in the case of malignancy. However, the presence of carcinomatous microfocuses without capsular invasion, and the absence of neighbouring invasion or metastases, means that surgical management can be limited to removal of the cyst. In all other cases, a total thyroidectomy combined with removal of the TTC and internal irradiation with radioactive iodine is required, with long-term monitoring. Complete, well-managed treatment is generally associated with a good long-term prognosis.

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