

CASE REPORTS

Papillary carcinoma of the thyroglossal duct cyst: report of two cases

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Abstract

Aim: To present two cases of papillary carcinoma of the thyroglossal duct cyst (TDC) and to report their management. **Patients and Methods:** Two patients, a 47-year-old woman and a 60-year-old man, were operated in 2006 and 2008 in the Department of Digestive and Metabolic Surgery of the "Jean Verdier" Hospital. **Results:** The diagnosis of papillary carcinoma was established after the pathological examination carried out on the thyroglossal duct cyst, after its complete surgical excision. A total thyroidectomy followed by a treatment by radioactive iodine and a substitutive hormonal therapy were carried out in the second time for one of the two patients. For the other patient, a total thyroidectomy followed by a substitute hormonal therapy, were achieved. The evolution was favorable, with a follow-up of four years for the first patient and two years for the second one. **Discussion:** Total thyroidectomy after the complete excision of the cyst is currently the recommended treatment for the papillary carcinoma of the TDC. It was shown that a latent thyroid cancer could develop even 15 years after the initial excision of the thyroglossal cyst. The treatment is curative in 95% of the cases. **Conclusions:** Current treatment of papillary carcinoma of the thyroglossal duct cyst is well codified, allowing an excellent prognostic.

Keywords: thyroglossal cyst, papillary carcinoma, thyroidectomy.

□ Introduction

The cysts of the thyroglossal duct (TDC) originate in a failure of regression of the thyroglossal duct and can be located in an area from the base of the tongue to the pyramid of Lalouette.

They are the most frequent congenital cervical anomaly that occurs in 7% of the adult population [1]. The incidence of carcinoma arising in a thyroglossal duct cyst occurs in about 1%. It is diagnosed mostly during the fourth decades of life. The majority of the cases are discovered during the pathologic examination of the surgical specimen [2]. To date, around 260 cases were described in the literature.

Two cases of papillary carcinoma developed on a TDC are reported.

□ Patients, Methods and Results

Case No. 1

Mrs. E.A., 47-year-old, without any particular medical antecedents, consulted for an anterior midline neck mass known for several years but lately increased in size and became symptomatic, without dysphagia or dysphonia. The clinical examination found a submental mass, measuring 4 cm in diameter, soft, painless, mobile with swallowing, without associated inflammatory signs, isolated, without cervical adenopathy. A cervical

ultrasound showed a heterogeneous hypoechogetic mass associated with cystic images, in median position between the thyroid cartilage and the hyoid bone; the aspect of thyroid gland being normal. In view of the diagnosis of symptomatic thyroglossal duct cyst, its surgical excision was carried out through a horizontal cervicotomy centered on the lesion.

Histopathologic examination showed a nodular mass of $3 \times 2 \times 1$ cm, with a cystic aspect of non-encapsulated tumor (Figure 1a) at the section. Microscopically, the solid lesion consisted of papillae (Figure 1, b and c) of thin connective-vascular axis, tapered by cubic-cylindrical cells, compact vesicles or compact clusters of tumoral cells (Figure 1d). The nuclei of the cells were discreetly irregular, with a nuclear membrane well visible (Figure 1c). Some of the cell nuclei were clarified. These morphological aspects were evocative of papillary carcinoma of thyroid type developed on a thyroid heterotopia.

After multidisciplinary discussion, the patient underwent a total thyroidectomy, without lymph node dissection, because of absence of adenopathy. The final pathology of the thyroidectomy specimen did not report any tumoral foci. Nevertheless, the patient received a postoperative treatment by radioactive iodine and hormonal thyroid suppression therapy.

After four years, the clinical, radiological and biochemical assessments of the patient are normal.

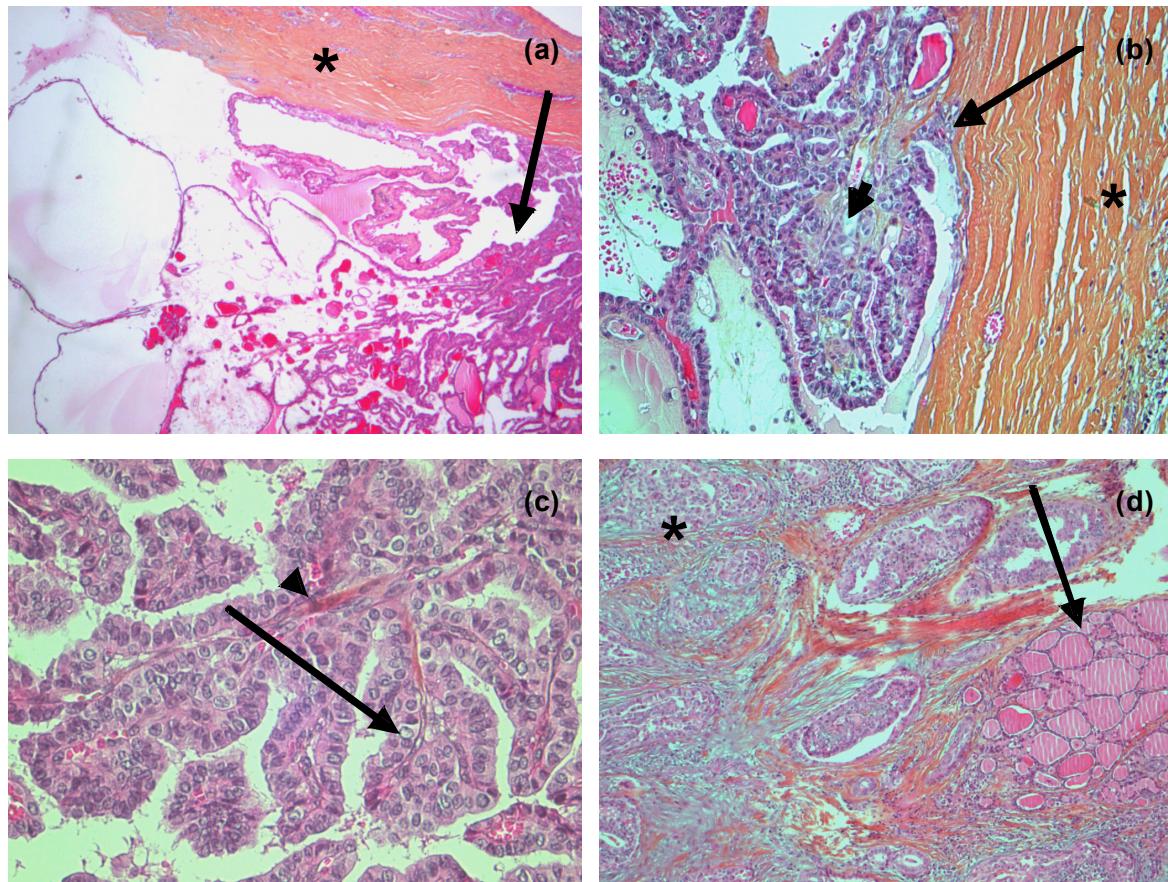


Figure 1 – Case No. 1. The nodule presented a cystic zone with fibrous walls (a, b: *) and intraluminal papillary projections (a, b: →). The papillae consisted of a connective-vascular axis (b, c: arrowhead). The epithelial cells were cubic, with clarified nuclei (c: →). Near this cystic zone the tumour was solid (d: *) and in contact with normal thyroid cells (d: →).

Case No. 2

Mr. S.J., 60-year-old, with associated morbidity of diabetes and the disease of Lapéryronie, consulted for the onset of a median submental cystic mass, with a diameter of 2 cm, mobile and painless. Preoperative ultrasonography showed a half-cystic and half-solid midline neck mass. The magnetic resonance imaging (MRI) of the base of the neck reported a median basal cervical mass, which corresponds to a cyst of the thyroglossal duct (Figure 2).

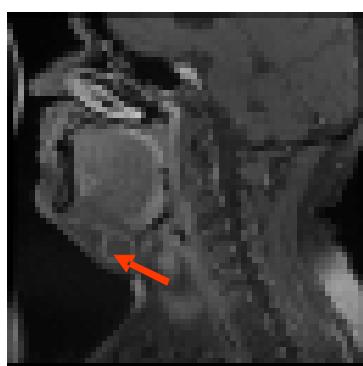


Figure 2 – Case No. 2. MRI of the TDC (arrowhead).

Surgical excision of the cyst was performed on day surgery. The surgical report described the presence of strong adherences between the cyst and the muscular layers.

The pathology examination reported a cystic mass measuring 2×2×1 cm, hemorrhagic on the section. Microscopically, within the cystic cavity, an epithelial proliferation of papillary architecture was identifiable. The immunohistochemical study was positive for anti-thyroglobulin and anti-TTF1. Cytology from the fluid within the cyst showed cells of epithelial allure with nuclear atypia.

A postoperative thyroid ultrasonography discovered three nodular lesions in the left lobe (one cystic lesion of 18 mm at the inferior pole and two tissular micro-nodules of 3 and 4 mm) and the absence of jugular carotid adenopathy. After discussion in multidisciplinary meeting, a total thyroidectomy without lymph node dissection was performed. The pathologic examination showed that the left thyroid nodule corresponded to a reshuffled vesicular adenoma, cystic and hemorrhagic, without identifiable carcinomatous lesions.

The patient did not receive any treatment by radioactive iodine. In the postoperative period it was performed a test with thyrogen which did not detect any residual thyroid cells.

Discussion

The papillary adenocarcinoma accounts for 75–85% of all the malignant tumors of the thyroglossal duct. Cancer on TDC does not have its own symptoms in 70% of the cases. Seldom, it can be revealed by

dysphagia, snores or loss of weight [3]. The average age of discovery is 40 years; the sex ratio female/male is 3:2 [2]. At the clinical examination, this lesion has the form of a median cervical nodule or a sinus opening with intermittent serous secretion in the area ranging between the hyoid bone and the cricoid cartilage [4].

Papillary carcinoma is the most frequent type (85%), insulated or associated with follicular forms (mixed variant). Epidermoid cancers are rare (7%) and it is exceptional to find carcinoma with Hurthle's cells, anaplastic carcinoma and association between papillary and epidermoid carcinoma [2, 4].

The three diagnostic criteria of papillary carcinoma on TDC are [5]: histological identification of the TDC (presence of thyroid follicles in the wall of the cyst), the existence of normal thyroid tissue adjacent to the tumor and the absence of primary carcinoma in the thyroid body. The origin of primary papillary carcinoma of the TDC [6] would be the presence of small islands of thyroid tissue adjacent to the cyst.

These carcinomas have a variable ultrasonographic aspect [4]: anechoic lesions, homogeneous or heterogeneous hypoechoic complex. The CT-scan and the MRI can be useful for the diagnosis: the presence of hypodense masses, calcifications (markers of papillary carcinoma [7]) which can be found on a simple cervical profile X-ray. The fine needle aspiration cytology can point out to malignancy, but the rate of true-positive is only 53% [8].

The CT-scan must be carried out for a solid cyst, deep-fixed and with associated adenopathy. The systematic ultrasonography of the neck should seek an associated thyroid pathology not detected by the clinical examination [9, 10].

The excision of the cyst, without thyroidectomy, remains valid only for the microscopic carcinomatous foci without invasion of the wall of the cyst. Total thyroidectomy, in the same operative time, if it is possible, followed by radiotherapy by radioactive iodine, is recommended by the majority of the authors [11, 12]. Kennedy TL *et al.* [11] point out that the patients treated for carcinoma on TDC should receive hormonal suppression therapy to prevent the thyroid tissue stimulation, tumoral or normal, by L-thyroxin.

In case of adenopathy discovered during the complementary examinations or in preoperative, a lymphatic dissection of jugular carotid lymphatic chain is indicated [13, 14].

The prognostic of the papillary carcinoma of the TDC is identical to that of papillary carcinoma of thyroid, with a rate of curability of 95% [9]. In the cases where the thyroidectomy was not carried out, a monitoring in the very long term is necessary to detect a latent thyroid cancer [12].

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Conclusions

The authors add to the literature two new cases of papillary carcinoma of the thyroglossal duct cyst. Papillary carcinoma on TDC is rare and the diagnostic is often histological, but ultrasonography of the neck is recommended in first line. In the case of histological confirmation, a CT scan is recommended.

The treatment of carcinoma of the thyroglossal duct is well codified today and allows an excellent prognostic.

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