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Micro-Papillary Carcinoma Arising from a Thyroglossal Duct Cyst: Case Report

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Abstract | **Case Report**

A thyroglossal duct cyst is the most common congenital cyst in the cervical region. Thyroid papillary carcinoma incidence in thyroglossal duct cysts is considered to be low. In most cases, the diagnosis of thyroglossal duct cyst papillary carcinoma is made postoperatively. We present a 53-year-old female patient with thyroid papillary carcinoma which developed from a thyroglossal duct cyst. This was confirmed in a histopathologic study after operation. In our case, there was neither lymph node involvement nor invasion of adjacent tissue. The patient then underwent total thyroidectomy along with the Sistrunk operation. The patient was followed up for 6 months without any metastasis or recurrence.

Keywords: Papillary Thyroid Carcinoma, Sistrunk Procedure, Thyroidectomy, Thyroglossal Cyst, Thyroglossal Duct Cyst Carcinoma.

INTRODUCTION

Pathology of the thyroglossal duct is one of the main differential diagnoses of midline anterior neck swelling. Thyroglossal duct cysts constitute the majority of these pathologies. They affect about 7% of the world's population [1, 2].

They constitute one of the most important differential diagnoses for a swelling of the midline anterior neck. Thyroglossal duct cysts (TDCs) make up the majority of these pathologies. They are found in about 7% of the world's population [1-3].

Thyroglossal carcinoma (TDCa) is rare. First-line radiological examination is ultrasound. Computed tomography (CT) or magnetic resonance imaging (MRI) may be indicated in the presence of atypical ultrasound features (i.e. solid components or calcification densities) or strong clinical suspicion of tumour, to document an orthotropic thyroid gland and to assess and characterise the features and extent of neoplastic processes [4].

Complications associated with thyroglossal duct cyst (TGDC) include cosmetic problems, fistula formation and recurrent infections. There is also a < 1% chance of developing TGDC carcinoma, most commonly papillary (92%) or other less common types such as squamous (5.2%) or follicular (1.7%) [5].

The most common surgical procedure for TGDC is the Sistrunk procedure, which involves the removal of the TGDC, the central part of the body of the hyoid bone and a core of tissue around the thyroglossal tract, which opens into the oral cavity at the cecal foramen [6].

We report a case of a classic papillary carcinoma arising in the TGDC, associated with papillary carcinoma arising in the thyroid.

CASE PRESENTATION

A 53 years old female patient was referred to our Maxillofacial Surgery Department at the Ibn Sina Hospital in Rabat with a median submental mass evolving for 6 months. The mass was slowly increasing in size with no dysphagia, no hoarseness and no dysphonia. There was no past history of recent upper respiratory tract infection or use of medication. The patient didn't have any history of neck irradiation or any clincal signs of hypo or hyper-thyroidism and no family history of thyroid cancers.

Physical examination revealed a single well-circumscribed, mobile, midline submental swelling of approximately 3 cm in diameter, that was moving with tongue protrusion and swallowing. The overlying skin had a normal appearance. Thyroid palpation was otherwise normal without cervical lymphadenopathy. The thyroid gland was clinically regular in shape and size. The rest of the systemic examination had no particularity.

Figure 1: Image showing midline submental swelling of approximately 3 cm of diameter

The patient underwent a cervical computed tomography which revealed cystic dilatation of the right Wharton's duct measuring 19 mm, enclosing a well-limited formation of discreetly irregular contours, tissue density containing some calcifications, with thickening of its wall, associated with multiple bilateral laterocervical and supraclavicular adenopathies with heterogeneous density. Thyroid gland had normal size and aspect.

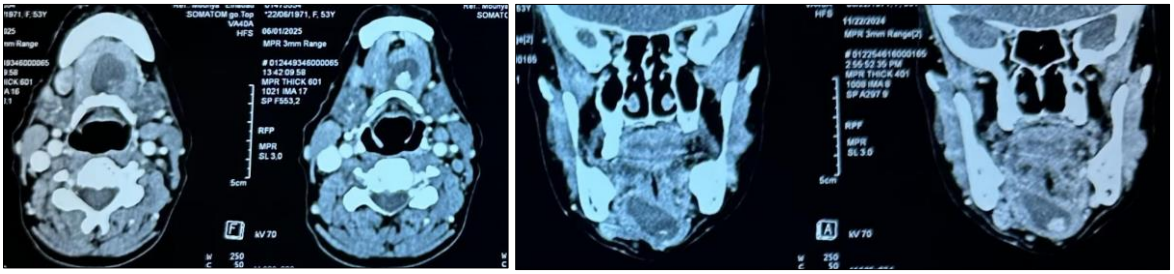

Figure 2: Neck CT showing cystic dilatation of the right Wharton's duct measuring 19 mm

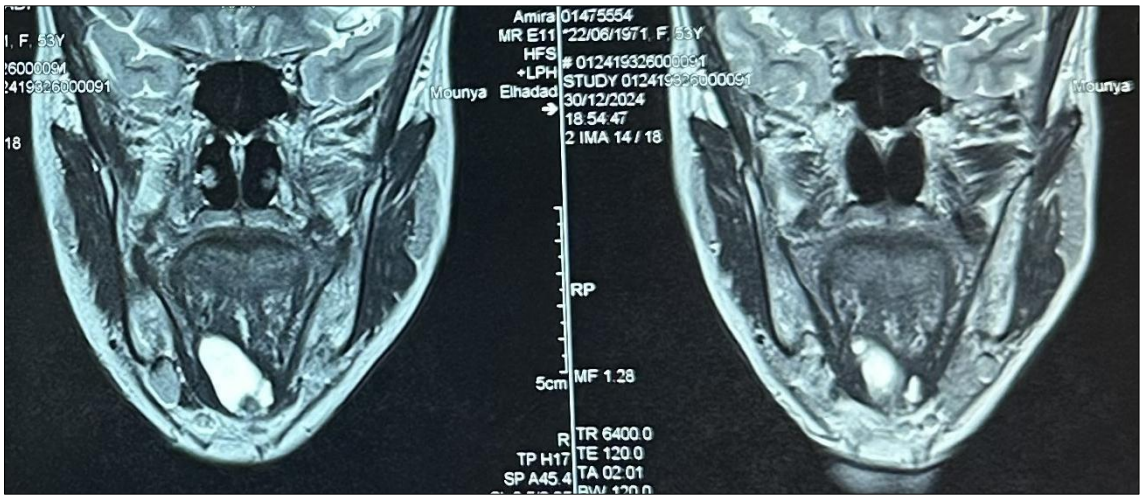

Figure 3: MRI showing dual-component under-chin mass

Figure 4: Operatory specimen

Following our multidisciplinary team, the patient underwent, under general anesthesia, a total surgical excision of the cyst and a tongue biopsy, using a double approach: floor of the mouth and midline cervicotomy. The patient had no post-operatory complications and the evolution was satisfying.

The histological examination of the surgical excision specimen showed papillary thyroid microcarcinoma on thyroglossal tract cyst.

The patient was then readmitted for a total thyroidectomy along with the Sistrunk operation. Later on, the patient was given adjuvant treatment with radioactive iodide and thyroid suppression therapy.

DISCUSSION

The thyroid is the earliest endocrine gland to develop in the human embryo [7]. It descends from the cecal foramen at the base of the tongue via the thyroglossal duct to the anterior part of the neck in close proximity to the hyoid bone. A remnant of the thyroglossal duct, most commonly a cyst, develops due to incomplete atrophy of the duct, usually by the 7th week of gestation [8]. TDC is the most frequent non-odontogenic cyst manifesting as a neck mass at any site along the tract [9].

We present a rare case of a submental mass presenting as a classic papillary carcinoma arising in the TGDC, associated with another papillary carcinoma arising in the thyroid gland. Thyroid tissue is normally located in the wall of the TGDC and has the potential to harbour malignant tumours, most commonly papillary thyroid cancer [10]. It's rare, slightly more common in women [11]. The cause of TGDC carcinoma is unclear. Theories include metastatic disease from an occult primary, or spontaneous development from ectopic thyroid tissue found within the wall of the TGDC [10].

Symptoms of TGDC carcinoma cannot be distinguished from benign TGDC. Therefore, a fast increasing growth with compression symptoms or the presence of a firm or hard, irregular mass may be signs of TGDC carcinoma [6-12].

The primary examination of a thyroglossal duct cyst should always comprise a complete physical examination of the head and neck region, with emphasis on the evaluation of the thyroid gland and lymph nodes, and thyroid function tests and further studies to establish the diagnosis, especially those related to the evaluation of functional thyroid tissue and its location, given the potential for finding ectopic thyroid tissue adjacent to the cyst as a single functional gland [13].

The average age reported in literature is usually ≥ 40 years old which is consistent with our case [14]. The female gender of our patient also underlines the finding that more women than men are affected by TGDC carcinoma [11]. The management pathway of TGDC carcinoma varies according to the histopathology. Preoperative evaluation should include a complete physical examination, head and neck examination, and thyroid evaluation (thyroid function tests, thyroid scan, biopsy). In our case, there was no clinical suspicion of malignancy. Pre-operative imaging showed no invasion of the thyroid gland or adjacent structures.

TGDC, branchial cleft cyst, lipoma, metastatic thyroid carcinoma, dermoid cyst, sebaceous cyst and enlarged lymph nodes are among the differential diagnoses of midline neck swelling [15].

The diagnosis of TGDC carcinoma is usually made intraoperatively or at definitive histopathology, which is the main difficulty. TGDC carcinomas should also be distinguished from papillary carcinomas arising from the apex of the pyramidal lobe [6].

The surgical procedure involves surgical excision of the TGDC (Sistrunk's procedure, the standard treatment involving removal of the entire ductus and part of the hyoid bone) and total thyroidectomy [7]. Some authors recommend total thyroidectomy in all cases of TGDC carcinoma, due to the high incidence of the presence of concurrent thyroid malignancy in the main thyroid as the pathophysiology; or to provide radioactive iodine therapy and adjuvant treatment therapy [6].

Others suggest total thyroidectomy as a standard procedure for TGDC carcinoma, however, the advantages of this are debatable based on the risks related to total thyroidectomy, such as a 1%-2% incidence of recurrent laryngeal nerve injury with redo surgery [16]. Sistrunk procedure can be performed as a single procedure in patients with a clinically and radiologically normal thyroid, with low risk, low malignant potential, patients younger than 45 years, without lymph node metastases, without previous irradiation, and with a negative surgical margin. Routine total thyroidectomy after TGDC carcinoma removal should not be performed in these patients. [14] Indications for total thyroidectomy include concomitant primary thyroid cancer, invasion of the cyst wall of the TGDC, and tumours > 1 cm in diameter [17].

In our case, the TGDC were excised. After histological examination, the patient was then referred for a total thyroidectomy. Radical neck dissection wasn't performed, due to the lack of evidence of cervical lymph node involvement.

Careful long-term follow-up is also important, as papillary carcinoma is usually a low- grade malignancy, and recurrences, if they occur, can be successfully treated with careful patient follow-up.

CONCLUSION

Thyroglossal duct cyst-associated with carcinoma is usually not suspected preoperatively. The management of these cases continues to be controversial due to the limited number of reported cases, so multidisciplinary management and individualization of each case play a fundamental role in the management of these rare cases. The Sistrunk procedure appears to be adequate for most of the patients, and only in some selected cases is total thyroidectomy or cervical lymph node dissection indicated.

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