

An Ectopic Papillary Carcinoma In A Thyroglossal Duct Cyst Leading To An Incidental Papillary Thyroid Carcinoma: Case Report

Benfadil Drissia*, Azedine .Lachkar, Chaymae Mezdid and Fahd El Ayoubi

Faculty of Medicine and Pharmacy Oujda, University Mohamed I oujda, ENT and Maxillo-facial department, University Hospital Center Mohammed VI, Oujda, Morocco

*Corresponding author:

Benfadil Drissia,

Faculty of Medicine and Pharmacy Oujda, University Mohamed I oujda, ENT and Maxillo-facial department, University Hospital Center Mohammed VI, Oujda, Morocco,

E-mail: benfadilrissia2@hotmail.fr

Received Date: 05 Feb 2023

Accepted date: 13 Mar 2023

Published Date: 18 Mar 2023

1. Abstract

Thyroglossal tract cyst (TTC) is the most common form of congenital anomaly of the thyroglossal tract. Papillary carcinoma on ectopic thyroid tissue in the thyroglossal tract is rare (less than 1%), and usually the diagnosis is made postoperatively as it clinically presents as TTC.

In this work we report a rare case of ectopic papillary thyroid carcinoma at the thyroglossal tract of a 38 years old woman, resected by the Sistrunk procedure. After diagnostic confirmation by histological examination a total thyroidectomy was done. This was followed by complementary treatment with ira-therapy.

2. Introduction

Thyroglossal tract cysts (TTCs) are a benign congenital neck anomaly that results from a defect in the obliteration of the thyroglossal duct during embryological development. The duct passes close to the hyoid bone during its descent, resulting in a close relationship between these two structures. Ectopic thyroid tissue in the thyroglossal tract is a common congenital anomaly, and most are benign. However, the possibility of malignancy is rare (1-2%) [1]. Ectopic thyroid tissue is frequently found in the GDC, but it is very rare that thyroid cancer develops from ectopic thyroid tissue in the GDC.

The management of papillary carcinoma on ectopic thyroid tissue in the thyroglossal tract is still controversial, with some opting for

total thyroidectomy in addition to the Sistrunk procedure and others recommending only the Sistrunk procedure [2].

Here we present the rare case of a 38-year-old patient who underwent Sistrunk surgery for anterior CDT, and histological analysis found an ectopic papillary carcinoma. All thyroid gland examinations were normal, however, we performed a total thyroidectomy and histological studies identified a concurrent papillary carcinoma. The patient was treated with radioactive iodide.

3. Case Presentation

We present the case of a 38-year-old woman, with no family history of thyroid malignancy and no history of neck irradiation, who presented to our department with a painless swelling in the anterior midline of the neck that had been progressively increasing in size for 2 years. Physical examination revealed a firm, painless, well-demarcated mass, approximately 4 cm × 1.5 cm, with a smooth surface, located in the midline of the neck, between the hyoid bone and the thyroid cartilage, with normal skin over the lesion (Figure 1).

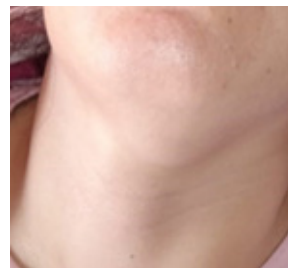


Figure 1: Midline cervical mass corresponding to thyrogloss cyst.

The mass moved with swallowing and tongue protrusion. The thyroid gland was of normal size and consistency and there were no palpable adenopathies.

Ultrasound revealed a supralaryngeal cyst 3 x 1.5 cm in diameter down to the hyoid bone, including solid tissue and calcifications within the cyst, which appeared to be a degenerated thyroglossal tract cyst (Figure 2).



Journal of Clinical Cases

Figure 2: Ultrasound showed a cyst containing a fleshy bud and parietal microcalcifications.

A cervical CT scan revealed a 32 mm midline cystic lesion in the region of the hyoid bone with tiny patches of calcification (Figure 3).

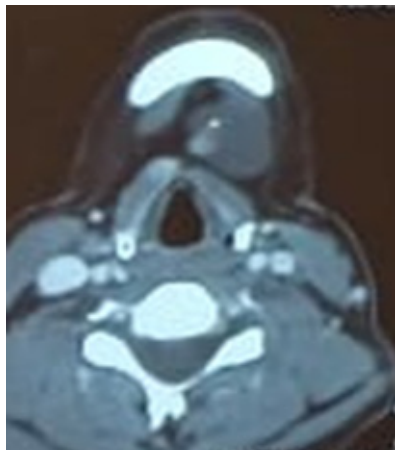


Figure 3: Cervical CT-scan with injection in axial section showing the thyroglossal cyst containing a fleshy bud and parietal microcalcifications and adhered to the body of the hyoid bone.

The patient underwent a Sistrunk procedure. Intraoperative findings revealed a 3 cm cystic swelling in the submental region that penetrated the body of the hyoid bone. The floor of the mouth muscles were adherent to the swelling. The tract was dissected to the base of the tongue with complete resection of the mass and tract.

Histopathological examination revealed fibrotic stroma, stratified squamous epithelium, and numerous true papillary structures that were lined with cuboidal cells without any evidence of wall invasion, leading to the diagnosis of papillary carcinoma on ectopic thyroid tissue in the thyroglossal tract (Figure 4).

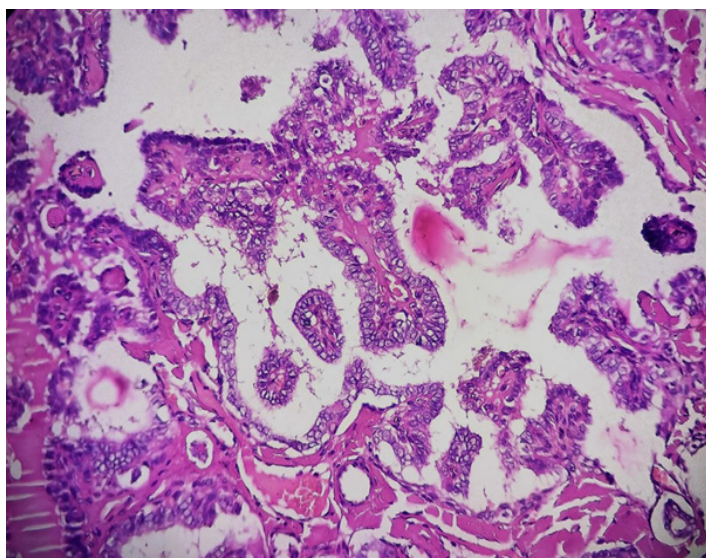


Figure 4: Tumor Has Papillary Architecture (*200, H&E).

After discussion of the case, a total thyroidectomy was performed 2 weeks later. Histopathological examination of the total thyroid specimen confirmed the diagnosis of papillary carcinoma without any evidence of wall or vessel invasion.

There were no complications in the postoperative period. Subsequently, the patient underwent radioactive iodine therapy.

4. Discussion

The thyroid gland descends from the foramen cecum of the tongue to the point below the thyroid cartilage at 7 weeks, after which an epithelial tract known as the thyroglossal tract is formed. Normally, this tract obliterates around the 5th-10th week of gestational life, but if it breaks through, it gives rise to a thyroglossal remnant, which may present as a cyst, tract, fistula, duct or ectopic thyroid tissue [1].

Ectopic thyroid tissue in the thyroglossal tract is a common congenital anomaly, and most are benign. However, the possibility of malignancy is rare (0,7 - 1%). The most common types are papillary carcinoma, follicular or mixed papillary-follicular carcinoma, squamous cell carcinoma and adenocarcinoma. Ectopic thyroid tissue in CDGT is a common finding, but its malignant transformation is a very rare anomaly [2].

On physical examination, TGDC may present as a small, painless, mobile swelling in the anterior neck. It is often asymptomatic, but may occasionally be inflamed due to infection. The clinical signs of CDT carcinoma may be similar to those of a benign cyst of the thyroglossal tract, it may present as a solitary cystic neck mass without thyroid abnormality or lymph node, as in our case, which often makes the preoperative clinical diagnosis difficult. [3].

The preoperative evaluation of a patient with SCD should include: a physical examination of the head and neck with palpation of the thyroid gland, thyroid function tests, ultrasound, CT scan and cervical magnetic resonance imaging. The purpose of these examinations is to confirm the diagnosis of atypical cases, to detect any possible malignant changes within the cyst, to differentiate thyroglossal duct cysts from other lesions and to identify the presence of functional thyroid tissue in the neck. However, the diagnosis relies on histological analysis of the surgical specimen because the clinical presentation and evaluation of patients with carcinoma of ectopic thyroid tissue is similar to that of patients with benign disease [4].

The surgical procedure used for a thyroglossal duct cyst is sistrunk's with removal of the thyroglossal duct cyst, part of the hyoid bone and the remaining portion of the duct. However, the main problem is the management of papillary carcinoma within the TDC. Some authors perform the Sistrunk procedure alone and others suggest adding a concomitant total thyroidectomy, especially for patients with a high risk of synchronous tumours [5].

There are different theories about the causes of TDG carcinoma, which means that even its management will be controversial. The literature is divided on this issue, with some authors preferring total thyroidectomy with neck dissection as an extension of the Sistrunk procedure because they believe that the thyroglossal duct is a natural pathway for the spread of occult thyroid cancer, and others are content with surgical excision of the cyst because they believe that squamous cell carcinoma arises directly in the thyroglossal duct cyst [6].

The prognosis of papillary carcinoma arising in a thyroglossal duct cyst is very good with a survival rate of 5 years.

Our case is that of an ectopic papillary carcinoma in a TGDC which leads to a simultaneous thyroid carcinoma. Despite the non-suspicious imaging of the thyroid by ultrasound and CT scan, we performed a total thyroidectomy in extension of the Sistrunk procedure after a consultation meeting, and the histopathological analysis of the thyroid showed papillary carcinoma which leads to the theory of spontaneous development from ectopic thyroid tissue with simultaneous thyroid carcinoma.

5. Conclusions

Papillary ectopic carcinoma in the thyroglossal tract is very rare and its treatment is controversial. From our observation, we conclude that total thyroidectomy combined with removal of the thyroglossal tract cyst containing malignant ectopic thyroid tissue is the ideal treatment, even when radiological examinations of the thyroid are normal.