Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Pathological Anatomy

Incidental Discovery of a Papillary Thyroid Carcinoma in a Thyroglossal Duct Cyst: Clinical Case

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DOI: https://doi.org/10.36347/sjmcr.2024.v12i10.021 | **Received:** 28.05.2024 | **Accepted:** 01.07.2024 | **Published:** 09.10.2024

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Abstract **Case Report**

The thyroid gland descends embryologically from the foramen to its anatomical location below the thyroid cartilage. It leaves behind an epithelial tract known as the thyroglossal tract; this tract usually disappears during the 5th to 10th week of gestation. Our patient is 52 years old, without notable history, who presented a slightly left-lateralized median cervical swelling of 5 cm with a long axis, fixed in superficial plane and mobile in deep plane. TTCs are the most common congenital anomalies in thyroid development, but carcinomas on TTC are extremely rare, with 90% of them arising from thyroid remnants. Papillary types represent 94% and less than 5% are of squamous origin. Their cause is unknown and there are no predisposing factors, neither clinical history nor physical examination can lead to a preoperative diagnosis. Surgery remains the main line of treatment, performing the Sistrunk procedure with or without total thyroidectomy in addition to excision of the cervical lymph nodes in some cases. In low-risk patients, Sistrunk operation might be sufficient, while in high-risk patients, a combined modality approach should be adopted.

Keywords: Thyroid, Carcinoma, Thyroglobulin, Surgery.

INTRODUCTION

The thyroid gland descends embryologically from the foramen to its anatomical location below the thyroid cartilage. It leaves behind an epithelial tract known as the thyroglossal tract; this tract usually disappears during the 5th to 10th week of gestation. Incomplete atrophy of the thyroglossal tract or retained epithelial cysts, however, creates the basis for the origin of the thyroglossal duct cyst (TTC). The inability of this tract to close predisposes to the formation of a thyroglossal cyst [1].

A TTC is the most common abnormality in the development of the thyroid gland [2]. It represents almost seventy percent of the median masses determined during childhood and 7% in adults [3]. Only 1% of thyroid carcinomas develop from a TTC lesion [4]. Brentano in 1911 and Uchermann in 1915 are recognized as being among the first to describe a neoplasm in a remnant of the thyroglossal duct; the median age of patients is generally 40 years and most are asymptomatic cited by Weiss and Orlich [5].

CASE PRESENTATION

Our patient is 52 years old, without notable history, who presented a slightly left-lateralized median cervical swelling of 5 cm with a long axis, fixed in superficial plane and mobile in deep plane. Cervical ultrasound revealed a hypoechoic cystic lesion measuring 48x38 mm, with a solid central zone of 12mm long axis.

A cystectomy was performed in the otorhinolaryngology department of our establishment and the sample was received the same day in the anatomy and pathological cytology laboratory.

Macroscopically, it is a cyst with a long axis of 4 cm attached to a bone fragment measuring 1.6x1 cm. When opening the cyst, a brownish hemorrhagic liquid is noted with a cystic wall with a thickness of 0.3 to 0.7 cm and the presence of a beige fleshy area filling almost the entire lumen measuring 2 cm in its long axis, of soft friable consistency, without calcifications or necrosis. [Figure 1]

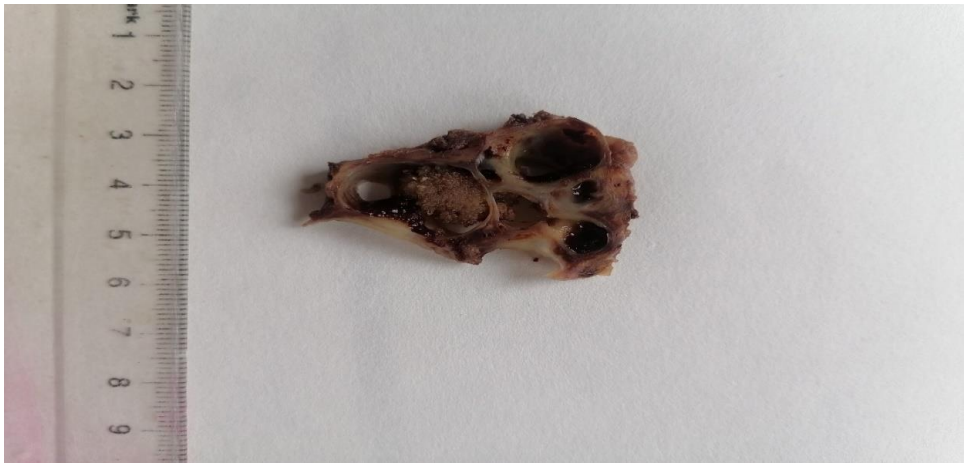

Figure 1: Macroscopic image showing the cross-section of the cyst resection specimen

Microscopically, it is a thick fibrous cystic wall bordered by an epithelium of the respiratory type in places and squamous in others, made of cells devoid of any cytonuclear atypia. The wall contains thyroid vesicles of variable size filled with colloid and lined with regular thyreocytes. The fleshy area is made up of a carcinomatous proliferation of papillary architecture [Figure 2], composed of cells with abundant eosinophilic cytoplasm and nuclei increased in size, with dense chromatin at the periphery, angular, grooved and incised with pseudo intranuclear inclusions and nuclear overlap. {Figure 3].

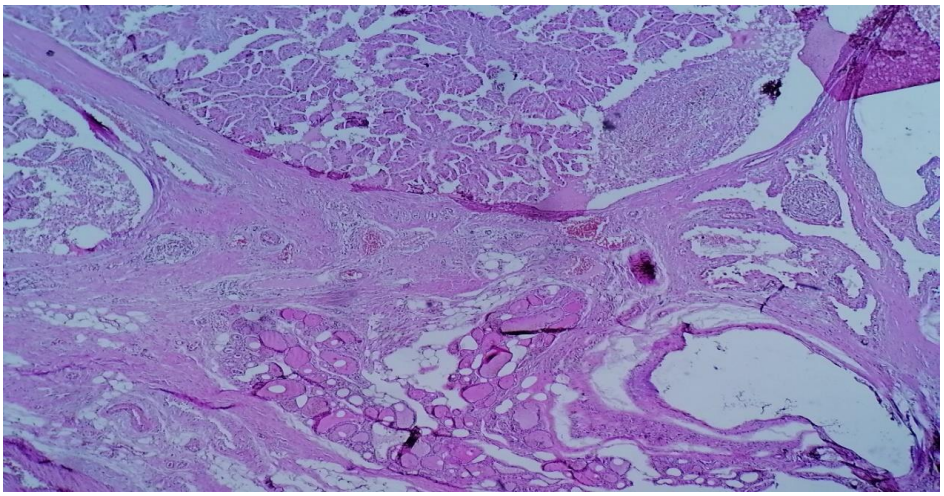

Figure 2: Histological image showing the presence at the level of the cystic wall of a papillary tumor proliferation depending on the thyroid parenchyma Gx10

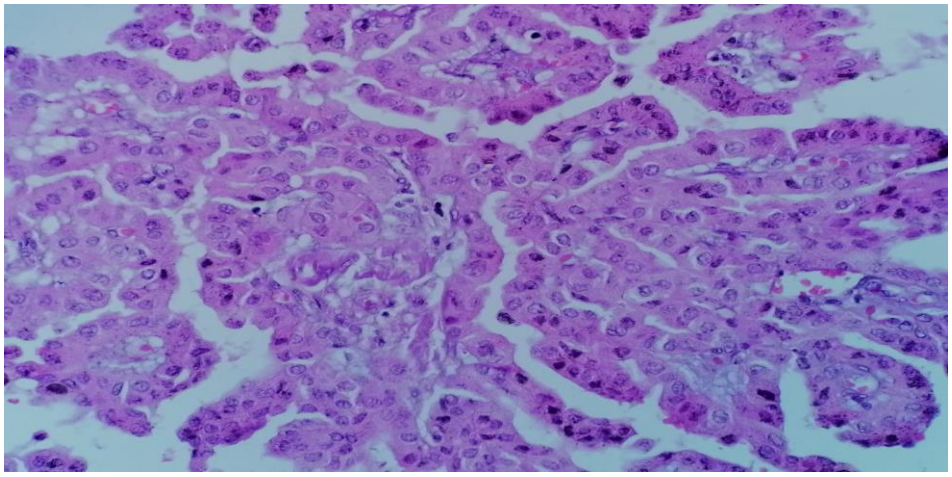

Figure 3: Histological image showing nuclear features of papillary carcinoma Gx20

An immunohistochemical complement was carried out and showed the positivity of the carcinoma cells for thyroglobulin [Figure 4], confirming the diagnosis of a papillary thyroid carcinoma developed on a cyst of the thyroglossal tract.

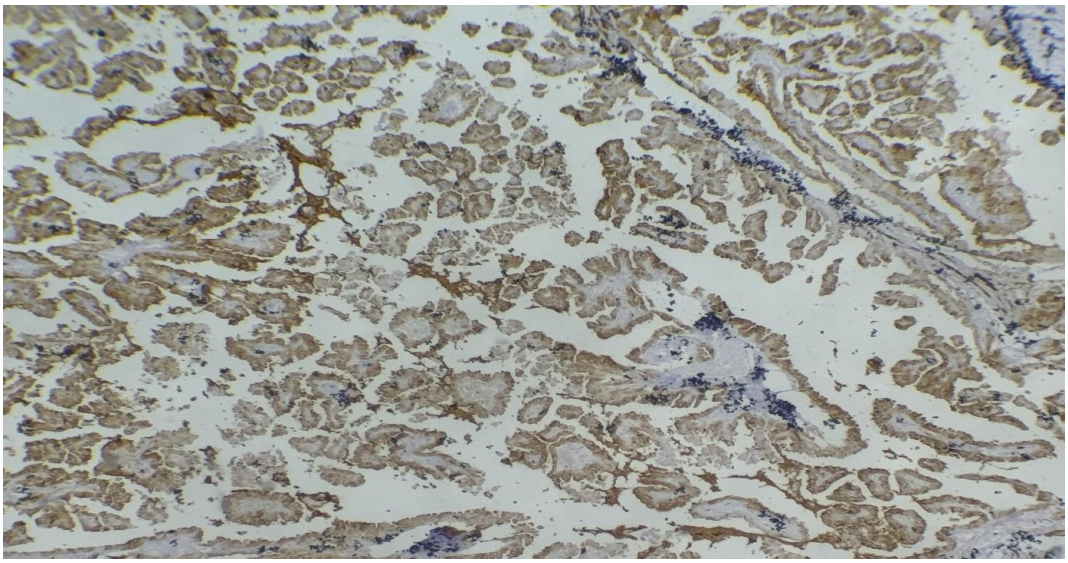

Figure 4: Immunohistochemical marking of tumor cells by anti-thyroglobulin antibody

DISCUSSION

KTTs are the most common congenital anomalies in thyroid development, but carcinomas on KTT are extremely rare, with 90% of them arising from thyroid remnants [5]. Papillary types represent 94% and less than 5% are of squamous origin [6]. Their cause is unknown and there are no predisposing factors, neither clinical history nor physical examination can lead to a preoperative diagnosis [7].

Generally, there are two theories to explain the thyroid origin of these carcinomas. First, the de novo theory is based on the fact that in 62% of cases, ectopic thyroid tissue can be identified histopathologically, which is supported by the absence of medullary carcinoma in KTT because it arises from parafollicular cells [8]. The second is the metastatic theory which suggests that thyroglossal cyst carcinoma is metastatic originating from an occult primary thyroid gland, as papillary carcinoma is multifocal in its nature [9]. Although Crile [10] believed that KTT could act as a natural channel for the spread of thyroid carcinoma, the metastatic theory seems less likely [11]. Mobini *et al.*, [12] admit that squamous cell carcinoma is probably the only true carcinoma of KTT, since other malignancies actually develop in ectopic thyroid tissue. Thus, squamous cell carcinoma can be considered the only primary tumor with a thyroglossal cyst, being very rare and with a poor prognosis with a mortality rate of 30 to 40% [13].

However, the diagnosis is often made postoperatively, upon histological examination of the resected section specimen. In one study, age of the cases ranged from 6 to 81 years, with a mean of 39 years. Women are affected more often than men [14]. To confirm a diagnosis of thyroglossecystic duct, the following criteria must be met: the cyst must be located in the mid-neck region; the wall of the cyst should be composed of cubic epithelial cells; and lymphatic tissues and normal thyroid follicles should be present in the cyst wall [15].

Malignant tumors developing from the roglossal duct have two origins: thyroid carcinoma arising from thyroembrionic remnants of the duct or cyst and squamous cell carcinoma arising from metaplastic columnar cells lining the duct. More than 200 cases of thyroglossal ductal carcinomas have been reported, in which papillary carcinoma accounts for 80% of cases, with the remainder being squamous cell carcinoma [16]. If calcification is seen internally, malignancy should be suspected. Calcification is the characteristic of papillary carcinoma in a thyroglossal duct cyst [17].

Imaging studies such as ultrasound, tomography, and nuclear magnetic resonance provide valuable information when findings results suggestive of malignancy are detected, such as invasion of neighboring structures, calcifications, or suspicious associated with lymphadenopathy, however, they do not do not in themselves represent a reliable diagnostic method [22].

The cytological examination obtained by FNA is only diagnostic in 66% of cases, as reported by Chala *et al.*, [21], with papillary carcinoma in 87% of cases, 5% squamous cell, 1.7% follicular and 0.9% anaplastic [18, 23]. In most cases, carcinoma originating from a thyroglossal cyst is treated with a resection with the Sistrunk procedure, with a reported 95% cure. In the Sistrunk procedure, the cyst, a part of the hyoid bone and the thyroglossal duct are removed together[19]. Some

studies argue in favor of total thyroidectomy in all patients due to the high risk of unsuspected infiltration with thyroid involvement from papillary thyroid carcinoma. Your roidectomy also allows for the addition of optional radioactive iodine treatment and more effective use of thyroglobulin levels for monitoring and follow-up [20]. The prognosis is excellent and reported distant metastases are very rare.

It has been found that regional lymph node metastases from primary papillary pillar carcinoma in thyroglossal duct cyst occur in only 7.7% of reported cases, and local invasion rarely occurs [26]. The risk of metastases is <2% of cases [24]. Our case showed the appearance of a unilateral reaction affecting the cervical lymph nodes but there was no distant spread.

Careful long-term follow-up of patients is important because even though papillary carcinoma is a low-grade malignancy, it is susceptible to recurrence which can be successfully treated during careful patient follow-up. All patients should be reassessed and undergo a neck CT scan every six months during the first year and annually thereafter [25]. Park said this patient's follow-up examinations consist of a thorough physical examination, ultrasound of the surgical area, thyroid, and whole body scintigraphy [27].

CONCLUSION

The current case report adds to published cases of primary thyroglossal duct cyst carcinoma; this is a rare tumor, but it should always be considered as an option when evaluating a neck mass. It is generally diagnosed postoperatively as an incidental finding on histopathological examination.

Surgery remains the main line of treatment, performing the Sistrunk procedure with or without total thyroidectomy in addition to excision of the cervical lymph nodes in some cases. In low-risk patients, Sistrunk operation might be sufficient, while in high-risk patients, a combined modality approach should be adopted.

Due to the rarity of this disease, there are currently no established evidence-based guidelines regarding the optimal surgical approach and subsequent management. Multidisciplinary team management should be considered to identify high-risk patients, who will require a more aggressive therapeutic approach.

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