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Thyroglossal duct cyst carcinoma: Case report

Carcinoma de cisto do ducto tireoglosso: Relato de caso

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ABSTRACT

Introduction: Considered the most common congenital anomaly of the development of the thyroid gland, the Thyroglossal Duct Cyst (DTC) has an estimated prevalence of 7% of the population, although the vast majority of these cysts are benign, about 1% of cases may malignancy. Objective: To report the history, evolution, and management of a patient with thyroglossal duct cyst who progressed to carcinoma, to review the pathophysiological character of the disease, and the scarce literature on this topic. Case summary: Female, 40 years old, diagnosed in 2018 with CDT. She seeks a Head and Neck surgeon in 2020 for reassessment of the case, to whom she reports an increase in the nodule and local discomfort. Surgery is indicated for its removal. The anatomopathological examination performed after surgery reveals suspicion of malignant transformation. Immunohistochemical examination confirmed the suspicion. A follow-up visit was requested in 6 months and ultrasound was requested to control the pathology. Discussion: The thyroglossal duct results in the permanence of the thyroid descent path from the base of the tongue to its final resting place in the anterior region of the neck in the midline. This tract should obliterate at the beginning of fetal life, failure in this involution can later lead to a cyst in this duct. The cyst is often noticed after infection of the upper respiratory tract and in rare situations, a carcinoma can develop from the walls of the cyst. Conclusion: cases of CDT carcinoma should be discussed due to the scarcity of reports in the literature and its rare incidence in the clinical field.

Keywords: Thyroglossal cyst, Congenital diseases and abnormalities, Carcinoma.

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INTRODUCTION

A thyroglossal duct cyst (DTC) is the most common form of congenital anomaly in developing thyroid. Approximately 70% of midline cervical masses in children and 7% of midline cervical masses in adults are thyroglossal duct cysts. Normally, during the third week of fetal development, the thyroid gland descends along the thyroglossal duct, a structure originating from the blind foramen of the tongue, through the base of the tongue toward the lower front of the neck, where it is normally found in adults (Figure 1). The thyroglossal duct physiologically disappears by the tenth week of gestation, in some cases it may fail to obliterate and form a cyst. CDT malignancy occurs rarely, only in about 1% of all cases, with papillary carcinoma being the most common type (1). CDT is usually asymptomatic and detected within the first two decades of life (2). A cyst is often noticed after infection of the upper respiratory tract, such as sinusitis, tonsillitis, otitis media, pharyngitis, and laryngitis, as this causes it to enlarge and become painful due to inflammation. In rare situations, a carcinoma can develop from the walls of the cyst. Carcinoma should be suspected if the cyst has the following characteristics: it is hardened, fixed, irregular, or associated with lymphadenopathy (3).

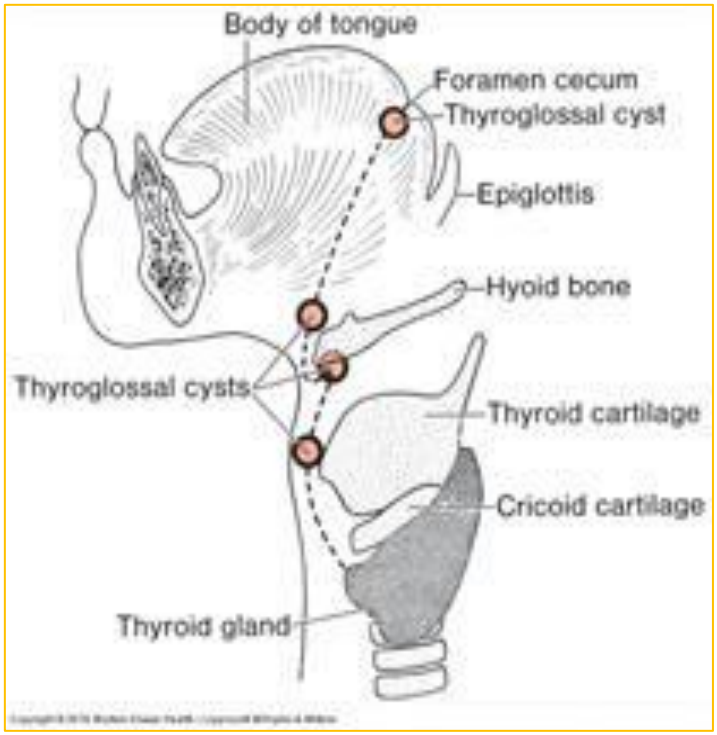
The pathological classification for malignancy occurring in thyroglossal duct cysts is divided into papillary carcinoma, which corresponds to 80% of the reported cases, follicular variant of the thyroid papillary which occurs in 8% of cases, squamous cell carcinoma in 6% and adenocarcinoma, anaplastic and follicular thyroid carcinoma correspond to 6%. In general, there is a predilection for female involvement. The mean age of occurrence is 39 years in DTC carcinoma of any type and 54 years in squamous cell carcinoma (7).

The evaluation of a patient with thyroglossal duct cyst should correlate the clinical findings with imaging tests: computed tomography (CT) or ultrasonography (USG) (8). In most of the cases reported in the literature, DTC carcinoma is only diagnosed as an incidental finding in the postoperative histopathological examination (8,9). Only a minority are diagnosed by fine needle aspiration (FNA) in the preoperative evaluation. Finally, the performance of anatomopathological examination in all cases is imperative for diagnostic confirmation(10).

For correct management, it is essential to differentiate between primary DTC carcinoma and metastatic carcinoma. The diagnosis of primary carcinomas originating from a DTC carcinoma is made based on three mandatory criteria: 1) presence of carcinoma in the DTC wall; 2) presence of normal thyroid gland tissue adjacent to the tumor; 3) presence of a clinically normal thyroid gland without any evidence of primary thyroid carcinoma. Sistrunk surgery and

thyroidectomy followed by ablation with iodine 131 and TSH suppression in metastatic lesions are indicated. Generally, the prognosis is favorable (20).

Figure 1. Migration path of the thyroid gland from the blind foramen of the tongue to its final position in the anterior portion of the neck.



Source: Embryology Learning Resource (Duke University Medical School)

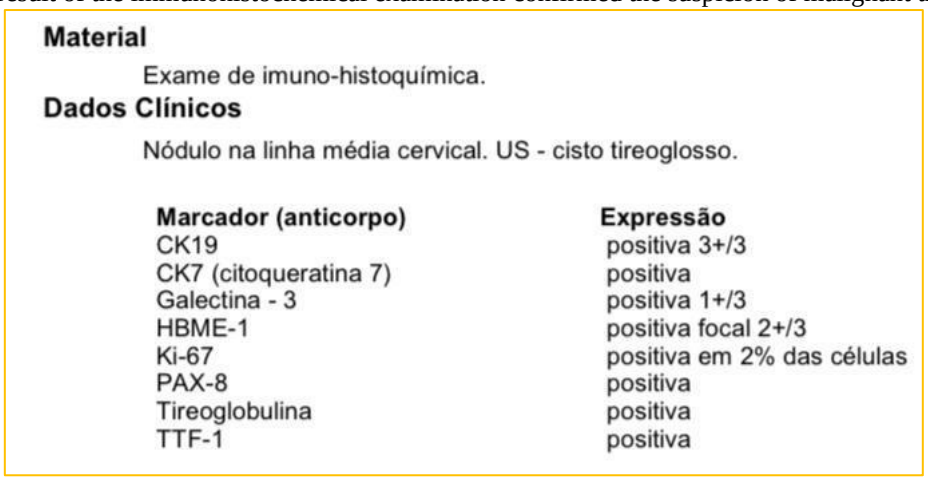
CASE REPORT

A 40-year-old female patient, healthy, non-smoker, non-alcoholic, with no history of allergies. In 2020, she sought a Head and Neck surgeon to whom she reported that 2 years ago she was diagnosed by an otorhinolaryngologist with a thyroglossal duct cyst. The patient reported seeking reassessment of the case due to the progressive increase of the nodule, with consequent local discomfort. The increase in volume was asymptomatic until then, slow-growing, and there was no history of similar cases in the family. As described in the literature, in our case, the patient started with a palpable, asymptomatic cervical mass.

After complementary examinations, the ultrasound showed a nodule of 2.4 cm, with a cystic aspect, well defined, suggestive of a thyroglossal cyst and the ultrasound performed in the surgeon's office revealed a thyroid of normal morphology and sizes, without nodules of any nature, cervical lymph nodes of usual appearance. After performing the necessary tests, surgery was indicated for its removal. The anatomopathological examination after surgical removal of the cyst revealed a thyroglossal duct cyst showing a focus with projection of an atypical papilla

on the wall, suspicious of malignant transformation, so the need for immunohistopathological examination was indicated (Figure 2); which confirmed the suspicion of malignancy: well-differentiated, classic, infiltrative papillary carcinoma in fibroadipose tissue. A six-month follow-up and an in-office ultrasound was requested to control the patient's evolution. The patient returns to the office asymptomatic, with no evidence of return of a palpable mass, confirmed by complementary tests to the physical examination.

Figure 2. The result of the immunohistochemical examination confirmed the suspicion of malignant transformation.



Material

Exame de imuno-histoquímica.

Dados Clínicos

Nódulo na linha média cervical. US - cisto tireoglosso.

Marcador (anticorpo)	Expressão
CK19	positiva 3+/3
CK7 (citoqueratina 7)	positiva
Galectina - 3	positiva 1+/3
HBME-1	positiva focal 2+/3
Ki-67	positiva em 2% das células
PAX-8	positiva
Tireoglobulina	positiva
TTF-1	positiva

DISCUSSION

The thyroid gland has an embryological origin near the blind foramen of the tongue, and passes through the developing hyoid bone, the thyroid descends through an epithelial canal known as the thyroglossal duct, this channel obliterates during the 8th and 10th gestational weeks. However, in incomplete atrophy of this duct, the remnant of the duct may form a cyst, pathway, fistula, or ectopic thyroid tissue in a cyst or duct. A thyroglossal canal cyst usually presents as a palpable asymptomatic midline tumor at or below the hyoid bone. Thyroglossal duct cyst is the most common non-odontogenic cyst, with a prevalence of 7% of the adult population (11).

A thyroglossal duct cyst carcinoma is a rare finding in the literature, occurring in less than 1% of thyroglossal duct cysts, the first description dates from 1911 and, until 2004, about 200 cases were described in the literature (14) having their origin in thyroid or epithelial tissue. The most common type is papillary carcinoma (80%), followed by mixed, papillary, and follicular carcinoma (8%), and squamous cell carcinoma (6%). The remaining 6% include Hürthle, follicular, and anaplastic cell carcinomas. (15). Squamous cell carcinoma arises from the metaplastic columnar epithelium (12). The clinical presentation is usually similar to that of

benign disease. Therefore, in most cases, the diagnosis of malignancy is made only after surgery (15). The incidence of papillary carcinoma arising in the thyroglossal duct cyst is <1% and is usually seen in younger women, with a sex ratio of 1.5:1 (13).

When there is clinical suspicion of malignancy, FNAB or imaging tests can aid in the diagnosis (14). Computed tomography (CT), when performed, shows a well-circumscribed, low-density, thin-walled lesion. In the case of malignancy, the most common findings are: a solid nodule in the cyst, followed by microcalcifications, irregular margin, and a thick wall (16). Microcalcifications may be a specific marker for papillary DTG carcinoma (17).

FNAB, although well indicated in suspected GTD cysts, is incapable of diagnosing follicular carcinoma. This shows the importance of an adequate clinical examination, which, associated with intraoperative findings, should be the basis of therapeutic decision in suspected cases(15). Since the absence of FNAB abnormalities does not exclude the presence of malignancy, especially if clinical suspicion is high, the possibility of finding a carcinoma should be discussed with patients prior to surgery (8).

Since carcinoma is most often an incidental finding after surgical excision, the criteria for diagnosis are: carcinoma must be in the DTG cyst wall; GTD should be distinguished from a lymph node cystic metastasis by histological demonstration of an epithelial line and normal thyroid follicles in the cyst wall; and there should be no malignancy in the thyroid gland or any other possible primary site. Often, the putative excised DTG cyst is the patient's only functioning thyroid tissue. However, an ultrasound image showing a thyroid gland in normal topography in patients with a presumed DTG cyst may exclude ectopic thyroid, but it is worth remembering that the specificity of the test is not 100%. Some authors suggest whether a scintigraphy investigation would be necessary to rule out ectopic thyroid tissue before submitting the patient to surgical treatment for a GTD cyst. However, scintigraphy is recommended only in two selected groups of patients: those with abnormal thyroid hormone levels and those in whom ultrasound cannot detect thyroid tissue in the neck (18).

In view of the persistence of the thyroglossal duct and cyst formation (with or without carcinogenic formation), the gold standard treatment so far is the Sistrunk technique, as it provides better results when compared to other techniques for the same purpose(5). There is a consensus that surgery for resection of thyroglossal duct carcinoma, or Sistrunk procedure, is the treatment of choice for "low-risk" carcinomas – less than 45 years of age, tumor smaller than 4 cm without extension to surrounding tissues, without distant metastasis – in the presence of a clinically and radiologically normal thyroid gland. This surgery consists of resection of the cyst

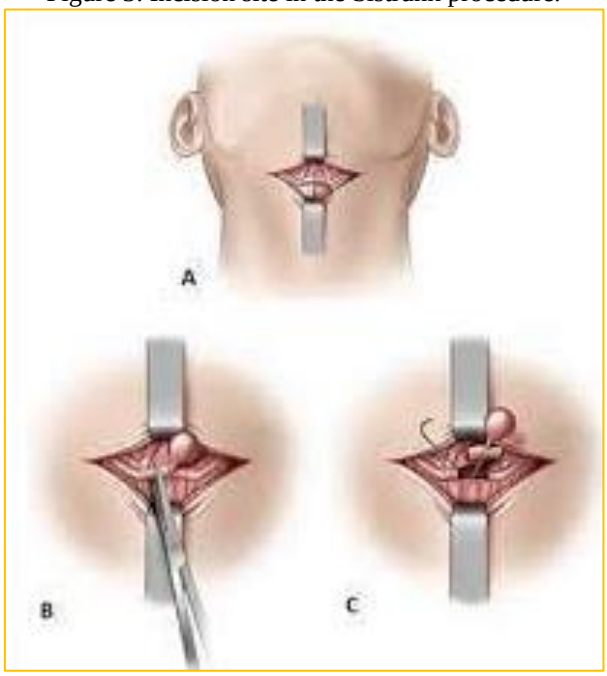
and duct, which extends to the blind foramen at the base of the tongue, in continuity with the middle portion of the hyoid bone (15).

On the other hand, those patients in the "high-risk" group – over 45 years of age, tumor larger than 4 cm, extending to the surrounding tissues, with lymph node metastasis or at a distance – require a broader treatment, including total thyroidectomy with or without neck dissection, followed by radioiodine therapy (19). In patients belonging to the "high-risk group", wider excisions should be made. This may include partial or total thyroidectomy when thyroid is involved, total laryngectomy, or neck dissection in case of distant metastases. Radiotherapy is indicated for cases in which there is a greater involvement, beyond the cyst wall, and is recommended in cases of total thyroidectomy (7).

The Sistrunk procedure consists of excision of the thyroglossal duct cyst, the central portion of the body of the hyoid bone, and a tissue nucleus around the thyroglossal tract to open in the oral cavity towards the foramen cecum (Figure 3). Consideration of adding thyroid resection in all patients is based on 3 aspects: (1) presence of thyroid malignancy, (2) use of radioactive iodine as adjunctive therapy, and (3) role of thyroglobulin as a follow-up marker. By using this procedure, the recurrence rate can be decreased significantly compared to simple excision: from 40% (simple excision) to 1-5% (Sistrunk procedure).

Based on the study by Balallaa et al., total thyroidectomy is indicated without considering the presence of thyroid gland involvement clinically or radiologically based on the premise that this procedure could aid in staging and detect metastases, and the risk of injury to the recurrent laryngeal nerve or parathyroid gland injury is considerably low, especially in the hands of an experienced operator (1).

Figure 3. Incision site in the Sistrunk procedure.



Source: Angélica S, et al. Thyroglossal cyst and Sistrunk operation. Rev Saúde Faciplac.

CONCLUSION

Carcinoma in thyroglossal duct cysts is a rare finding, with a predilection for females, with varied classifications, papillary being the most common, but in any case, it has been little reported in the literature. Thyroglossal duct cyst carcinoma develops from the cyst walls, which in itself is a rare finding in the population. The importance of describing their cases is due to the rare incidence in the clinical field and the need to know and suspect such pathology in the care of patients, who are most often asymptomatic, but attention should be paid mainly to hardened, fixed, irregular lesions associated with lymphadenopathy. Clinical suspicion is given through a good physical examination, which can be enriched with the adjunct of imaging tests such as ultrasonography and tomography, in addition to FNAB. The treatment of thyroglossal duct cysts, regardless of their malignant transformation or not, is always surgical, using the Sistrunk technique as the gold standard, and should be accompanied by anatomopathological examination

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