

## Thyroglossal duct carcinoma: Case report

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### ABSTRACT

**Introduction:** Although thyroglossal duct cyst carcinoma is a relatively easy pathology to treat, it is an uncommon finding that has been little reported in the literature. **Objective:** To report the history, evolution and management of a patient with a thyroglossal duct cyst who progressed to carcinoma, to review the pathophysiological nature of the disease, and the scarce literature on this topic. **Case summary:** Woman, 40 years old, diagnosed in 2018 with thyroglossal duct cyst. She seeks a Head and Neck surgeon in 2020 to evaluate 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. Immunohistochemistry 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 cyst carcinoma in the thyroglossal duct 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.

### INTRODUCTION

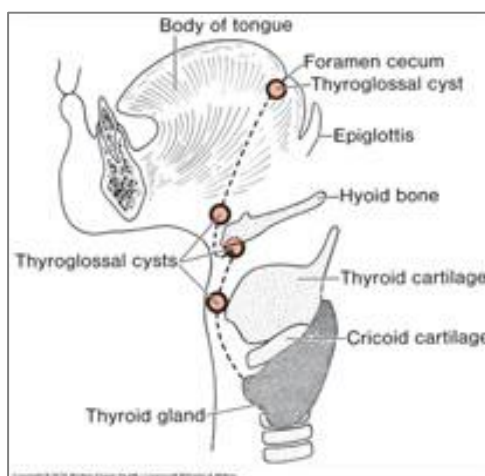
A thyroglossal duct cyst is the most common form of congenital anomaly in developing thyroid. Approximately 70% of midline cervical masses in children and 7% of midline cervical masses in adults are thyroglossal duct cysts. Normally, during the third week of fetal development, the thyroid gland descends along the thyroglossal duct, a structure originating from the foramen cecum of the tongue, through the base of the tongue toward the lower front of the neck, where it is normally found in adults (Figure 2). The thyroglossal duct physiologically disappears in the tenth week of gestation. In some cases, the thyroglossal duct may fail to obliterate and form a thyroglossal duct cyst. Thyroglossal duct cyst malignancy occurs rarely, only in less than 1% of all cases, with papillary carcinoma being the most common type (Diani K, et al. Case Report - Sistrunk Procedure on Malignant Thyroglossal Duct Cyst. Case Rep Oncol Med. 2020). The thyroid migration tract is called the thyroglossal duct, which normally atrophies, but when viable epithelium persists (7 to 41%) at some point during the migration, the thyroglossal duct cyst may occur, which corresponds to 70% of congenital lesions in the head and neck, and is usually asymptomatic and detected in the first two decades of life (Elisa BD, et al. Primary papillary carcinoma of the thyroglossal cyst duct – Case Report. Rev Salusvita, Ciên Biol Saúde (Bauru).

2020; 39(1):111-117.). 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 (Rosana LM, et al. Carcinoma of the thyroglossal duct. Rev Bras Cir Craniomaxilofac. 2012:127-129.). She is female-prone, with a mean age of 40 years. The clinical presentation of thyroglossal duct cyst carcinoma is similar to benign cysts. The main objective is to report the case of a patient diagnosed with thyroglossal duct cyst that malignant into carcinoma, a pathology of great relevance due to its low incidence.

Figure 1. The result of the immunohistochemical examination confirmed the suspicion of malignant transformation.

<b>Material</b>	
Exame de imuno-histoquímica.	
<b>Dados Clínicos</b>	
Nódulo na linha média cervical. US - cisto tireoglosso.	
<b>Marcador (anticorpo)</b>	<b>Expressão</b>
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

Figure 2. 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

The patient was a 40-year-old woman, healthy, non-smoker, non-alcoholic, with no history of allergies. Diagnosed in 2018 by an otolaryngologist with thyroglossal duct cyst. In 2020, she sought a

head and neck surgeon for reassessment of the case, to whom she reported local discomfort due to the progressive enlargement of the nodule. After ultrasound, which showed a 2.4 cm cystic, well-defined nodule suggestive of a thyroglossal cyst, surgery was indicated for its removal. Ultrasound performed in the surgeon's office revealed the thyroid of normal morphology and sizes, without nodules of any nature, cervical lymph nodes of usual appearance. The anatomopathological examination revealed a thyroglossal duct cyst showing a focus with projection of an atypical papilla on the wall, suspicious of malignant transformation, the need for immunohistopathological examination was indicated; which confirmed the suspicion of malignancy, a 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.

## **DISCUSSION**

The thyroid gland has an embryological origin near the blind foramen of the tongue, and passes through the developing hyoid bone. The thyroid travels down an epithelial canal known as the thyroglossal duct. This canal obliterates during the 8th and 10th gestational weeks. However, in incomplete atrophy of this canal, cysts and/or accessory thyroid tissue may appear in its remnants.

The remnant of the thyroglossal duct may be a cyst, a duct, a pathway, a 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. A thyroglossal duct cyst carcinoma is a rare finding in the literature, occurring in less than 1% of thyroglossal duct cysts, having its origin in thyroid or epithelial tissue. The most common type is papillary carcinoma, followed by mixed, papillary, and follicular carcinoma, and squamous cell carcinoma (Rosana LM, et al. Carcinoma of the thyroglossal duct. *Rev Bras Cir Craniomaxilofac.* 2012:127-129.). Because of its similarity to a benign cyst, the diagnosis of malignancy is made only after surgery. There is a consensus that surgery for resection of thyroglossal duct carcinoma, or the Sistrunk procedure, is the treatment of choice for "low-risk" carcinomas, patients aged <45 years, who have no history of radiation exposure, tumor size <4.0 cm, no soft tissue invasion, no distant or lymphatic metastases, and absence of aggressive tumor histology.

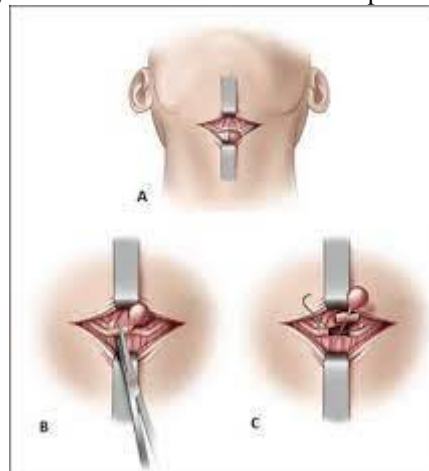
In view of the persistence of the thyroglossal duct and cyst formation (with or if carcinogenic formation), the gold standard treatment so far is the Sistrunk technique, as it provides better results when compared to other techniques for the same purpose (Angélica S, et al. Thyroglossal cyst and Sistrunk operation. *Rev Saúde Faciplac.*).

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). There are four approaches regarding surgical treatment for thyroglossal duct cyst malignancy, which are Sistrunk procedure alone, Sistrunk procedure with thyroid

lobectomy or pyramidal lobe resection, Sistrunk procedure with total or near-total thyroidectomy in all patients, and Sistrunk procedure with selective thyroidectomy for high-risk patients, male patients, patients aged 45 years or older, tumor size greater than 4 cm, presence of extracystic invasion, presence of lymph node metastasis, previous history of radiation, especially in the neck region, and presence of cold nodules on thyroid gland imaging. 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 (Diani K, et al. Case Report - Sistrunk Procedure on Malignant Thyroglossal Duct Cyst. Case Rep Oncol Med. 2020).

Figure 3. Incision site in the Sistrunk procedure.



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

## CONCLUSIONS

Carcinoma in thyroglossal duct cysts is a rare finding and, consequently, little reported in the literature. The importance of describing their cases is due to the rare incidence in the clinical field. The treatment of thyroglossal duct cysts, regardless of their malignant transformation or not, is always surgical.



## REFERENCES

- Diani K, et al. Case Report - Sistrunk Procedure on Malignant Thyroglossal Duct Cyst. *Case Rep Oncol Med.* 2020;ID 6985746. Disponível em: <https://www.hindawi.com/journals/crionm/2020/6985746/>.
- Elisa BD, et al. Carcinoma papilífero primário de ducto do cisto tireoglosso – Relato de Caso. *Rev Salusvita, Ciên Biol Saúde (Bauru).* 2020;39(1):111-117. Disponível em: [https://secure.unisagrado.edu.br/static/biblioteca/salusvita/salusvita\\_v39\\_n1\\_2020/salusvita\\_v39\\_n1\\_2020\\_art\\_09.pdf](https://secure.unisagrado.edu.br/static/biblioteca/salusvita/salusvita_v39_n1_2020/salusvita_v39_n1_2020_art_09.pdf).
- Rosana LM, et al. Carcinoma do ducto tireoglosso. *Rev Bras Cir Craniomaxilofac.* 2012:127-129. Disponível em: <http://www.abccmf.org.br/cmf/Revi/2012/julho-setembro/4-Carcinoma%20do%20ducto%20tireoglosso.pdf>.
- Florinda C, et al. Carcinoma papilar do canal tireoglosso – Relato de caso e revisão da literatura. *Rev Portuguesa Cir.* 2019;45:25-30. Disponível em: <file:///C:/Users/USU%20C3%81RIO/Downloads/707-1-2300-1-10-20191228.pdf>.
- Angélica S, et al. Cisto tireoglosso e operação de Sistrunk. *Rev Saúde Faciplac.* Disponível em: <http://www.roplac.com.br/revistas/index.php/RSF/article/view/336/186>.
- Michael A, et al. Invasive Thyroglossal Duct Cyst Papillary Carcinoma: A Case Report and Review of the Literature. *Am J Case Rep.* Available from: <https://www.amjcaserep.com/download/index/idArt/907313>.