BJO
Brazilian Journal of Oncology

CASE REPORTS ONCOLOGICAL SURGERY

Papillary carcinoma of the thyroglossal cyst as a metastasis of microcarcinoma of thyroid cancer: a case report and literature review

Carcinoma papilífero do cisto tireoglosso como metástase de microcarcinoma de câncer de tireoide: relato de caso e revisão da literatura

Sergio Altino Franzi[1], Tania Regina de Bastos Souza[1]

ABSTRACT

Papillary carcinoma of the thyroglossal cyst is clinically like the benign lesion in adults aged 15-45 year, with a higher incidence in females. It is a congenital anomaly of the midline of the neck and have the clinical worsening related to infections of high aerodigestive routes. We discuss clinical assess, diagnosis, surgery, hormonal suppression, radioiodine therapy, and prognosis in a patient with a complaint of nodule in the neck, and to the ultrasonography an image suggestive of the thyroglossal cyst without alteration of thyroid gland. He was submitted to Sistrunk's surgery and the anatomopathological exam revealed papillary carcinoma, and we do not find normal thyroid follicles in the thyroglossal cyst wall. We perform the total thyroidectomy by the thyroid alterations, showing in the anatomopathological exam a thyroid microcarcinoma. We completed by hormonal suppression and radioiodine therapy. The prognosis is favorable and the patient presented asymptomatic until now.

Keywords: Thyroglossal cyst; Thyroid Cancer, Papillary; Diagnosis; Combined modality therapy.

1. Hospital Geral Dr. José Pangella - Vila Penteado, Clínica de Cirurgia de Cabeça e Pescoço - Sao Paulo - SP - Brazil.
Financial support: none to declare.
Conflicts of interest: The authors declare no conflict of interest relevant to this manuscript.
Correspondence author: Sergio Altino Franzi.
E-mail: franzi.seal@gmail.com

Received on: February 22, 2021 | **Accepted on:** July 30, 2021 | **Published on:** August 16, 2021
DOI: https://doi.org/10.5935/2526-8732.20210023

RESUMO

O carcinoma papilífero do cisto tireoglosso é clinicamente semelhante à lesão benigna em adultos de 15 a 45 anos, com maior incidência em mulheres. É uma anomalia congênita da linha média do pescoço e tem a piora clínica relacionada a infecções de vias aerodigestivas altas. Discutimos a avaliação clínica, diagnóstico, cirurgia, supressão hormonal, radioiodoterapia e prognóstico em um paciente com queixa de nódulo no pescoço e à ultrassonografia uma imagem sugestiva de cisto tireoglosso sem alteração da glândula tireoide. Foi submetido à cirurgia de Sistrunk e o exame anatomopatológico revelou carcinoma papilar, e não encontramos folículos tireoidianos normais na parede do cisto tireoglosso. Realizamos a tireoidectomia total devido às alterações tireoidianas, evidenciando no exame anatomopatológico um microcarcinoma de tireoide. Completamos o tratamento com supressão hormonal e terapia com radioiodo. O prognóstico é favorável e a paciente apresentasse assintomática até o momento.

Descritores: Cisto tireoglosso; Câncer de Tiróide Papilífero; Diagnóstico; Modalidade de terapia combinada..

INTRODUCTION

The cyst of the thyroglossal duct comprises the congenital abnormalities that originate from the remains of the thyroglossal duct, the remaining ones being present in 7% of the adult population.[1] This duct originates from the pharyngeal apparatus, formed from a thickening of the primitive pharynx endoderm 24 days after fertilization, and is also known as primary thyroid gland. With the embryological growth of the tongue, this primordium descends to the neck region and there acquires its definitive shape in the 7th week of embryonic development.[2] If there is a remnant of this duct at any point along the path followed by the duct during the descent of the thyroid, there is formation of the thyroglossal duct cyst, which due to its embryonic origin, may contain thyroid tissue.[3] Although the occurrence of thyroglossal duct cyst is common, the development of cancer in the cyst wall is rare occurring in less than 1% of cases.[1]

Thyroglossal cyst is generally asymptomatic, and its presentation is similar, to cases of thyroglossal cyst benign, with the characteristics of a cervical mass in the hyoid region, asymptomatic, of cystic consistency, painless on palpation and mobile to swallowing and tongue protrusion, which are the findings in 70% of the occurrences and symptoms of dysphagia, dysphonia, and weight loss.[2]

Bhagavan et al. (1970),[4] reviewed 40 cases and found only three cases of painful mass and six cases of hardened mass. Joseph and Komorowski (1975)[5] studied 43 cases and found a very variable duration of evolution, with a median of one year and an average of 4.7 years, with extremes of 10 days and 40 years. There is a predominance of females over males of 1.5:1 in thyroglossal cyst carcinoma, with peak incidence for women and men in the third and sixth decade of life, respectively. [1] The preoperative evaluation consists of clinical examination of the head and neck, with palpation of the cyst, thyroid gland, and lymph node chains, in addition to the measurement of thyroid hormones and neck ultrasound.[3] The diagnosis is rarely made before surgery and should be suspected if a nodule is hardened, fixed or irregular at the level of the hyoid bone is found on palpation, which has recently changed its appearance, or when there is satellite lymph node enlargement. A rapid increase in the volume of the cyst may indicate malignancy; however, this also occurs when there is an acute spontaneous infection of the thyroglossal cyst.[3]

Katz and Hachigian (1988)[6] described a case diagnosed preoperative by cytology, however, the role of fine needle aspiration biopsy (FNAB) in the preoperative diagnosis has not been established in thyroglossal cyst carcinoma, given the difficulty of obtaining enough material to cytology, since the tumor is confined to the cyst wall. According to Hilger et al. (1995),[7] the diagnosis is a finding in the postoperative anatomopathological examination, with surgery being the ideal treatment for thyroglossal cyst carcinoma, which is the same for benign cyst, and consists of surgery with resection of the lesion by the Sistrunk operative technique.[8] This technique consists of an elliptical incision between the hyoid bone and thyroid cartilage, just below the cyst, which is dissected from the deep plane, pulled by the base, with excision proceeding towards to the base of the tongue, including the central portion of the hyoid bone and a lingual-based muscle cone.[8]

CASE REPORT

CLX, 42-years-old, male, Caucasian, married, taxi driver, from Sao Paulo. He sought to the head and neck service of General Hospital Dr. José Pangella – Vila Penteado on 01/21/2016, with a complaint of "nodule in the neck" 12 months ago with symptoms of local discomfort and discomfort when swallowing. He denies smoking and mentions social drinking. He denies arterial hypertension and diabetes mellitus. He denies toxophiliac. In the loco-regional

examination, a mass of four centimeters in diameter was observed, deep, in a midline, left antero-lateral infrahyoid, of hardened consistency, painless, mobile to swallow, without compromising the skin that covers it (Figures 1 and 2).

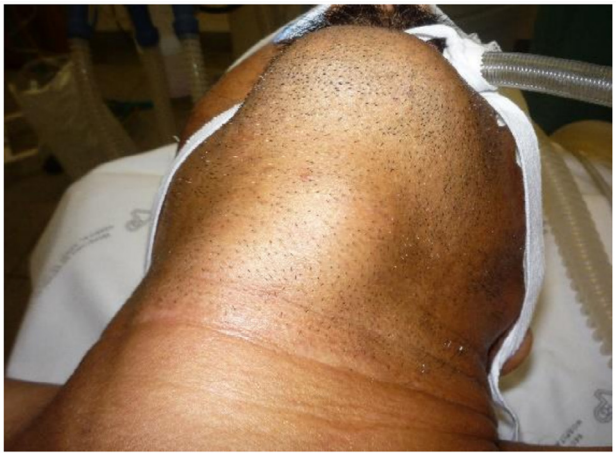

Figure 1. Photography of anterior nodular lesion of neck

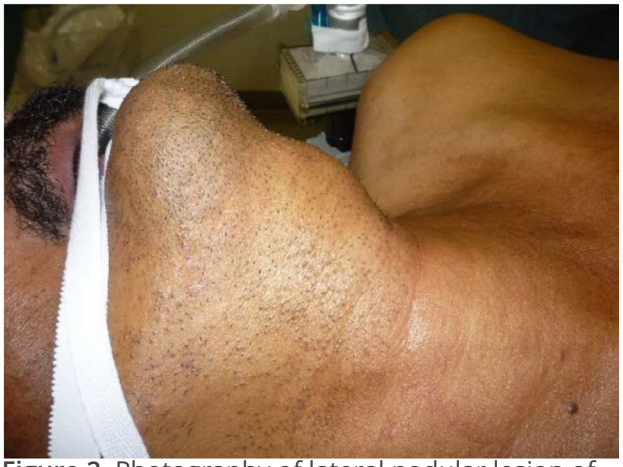

Figure 2. Photography of lateral nodular lesion of the neck

Preoperative exams revealed chest radiography without alteration, neck ultrasound, nodular image of the submental region, measuring 4.0cm x 4.0cm, with mixed echogenicity of a thyroglossal cyst, and with aspiration cytology of this nodular lesion that showed no malignant. In the loco-regional evaluation of the neck in the thyroid region, no thyroid nodule was palpated, also confirmed by the findings by computed tomography of the neck, with the remaining hematological, biochemical, coagulation, hormones, and thyroid antibodies unchanged. Direct laryngoscopy showed no changes.

The patient underwent surgical treatment on 04/26/2016 using the Sistrunk operative technique (Figure 3).[8] Upon returning, after 30 days, he reported pain at the surgery site and a feeling of discomfort in his throat and when swallowing, with "nervousness" due to the diagnosis of "tumor of the thyroglossal cyst" to the freezing test performed during the surgery. The anatomopathological exam in 06/2016, showed papillary carcinoma of classic pattern, measuring 1.8cm, histological and nuclear grade 2, not encapsulated, originating in a thyroglossal cyst, and infiltrating the connective tissue. Neoplasm-free margins, mitotic index of 2-4, undetected vascular and lymphatic invasion, undetected peri-neural infiltration and necrosis and hemorrhage present in about 10% of the tumor. The hyoid bone was free of neoplasia.

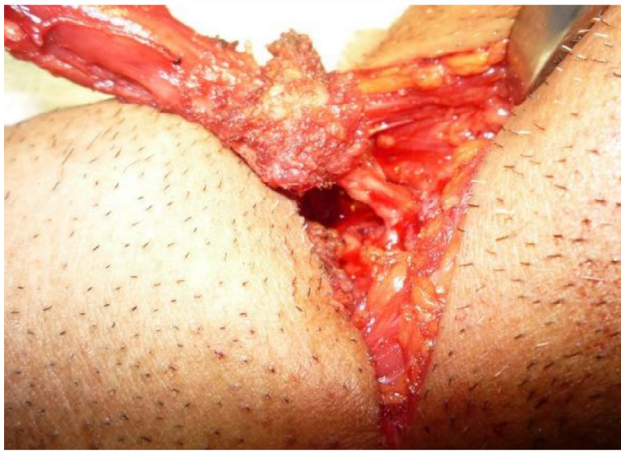

Figure 3. Photography of the surgical treatment of the thyroglossal cyst.

In view of these findings and the patient's report, we performed a new investigation of the neck with Doppler ultrasonography and found in the thyroid a nodule on the right measuring 0.2cm and on the left measuring 1.9cm, with aspiration cytology of a nodule on the left being performed negative for neoplasia and thyroid hormones tests within the normal range. In assessing the general conditions, prognostic, and demographic factors, it was offered to undergo total thyroidectomy and we did not recommend medication for hormonal suppression with sodium levothyroxine before surgery. The patient requested a reflection period of thirty days for this procedure, which underwent total thyroidectomy on 05/09/2016, without complications, and the intraoperative finding was the presence of nodular lesion in an irregular pyramidal lobe, hard measuring 0.6cm x 0.5cm nodular lesion in the right lobe measuring 0.2cm and in the left lobe the presence of a nodule greater than 1.9cm.

The patient evolved asymptomatic and the anatomopathological examination was diagnosed with papillary microcarcinoma of the right lobe of the thyroid measuring 0.2cm associated with adenomatous goiter of the left lobe, with negative neoplastic embolization, neoplasia-free thyroid capsule, and foreign body granuloma in the pyramidal lobe region. In view of these findings and associated with prognostic factors (42 years old, male, alcoholism, thyroglossal duct cyst size), being initially classified as low-risk, and based on the treatment protocol for these criterions, we performed total thyroidectomy,

based on the nodules thyroid disease found. In view the diagnosis of papillary microcarcinoma, as well as, in our conduct regarding a metastasis of papillary carcinoma in a thyroglossal cyst, we recommend radioiodine therapy with a total dose of 131mCi (03/2017) and with hormonal suppression with sodium levothyroxine 150mcg/day and calcium carbonate D3, 1g pills/day. There have been no complications to date and being followed up with the endocrinologist. We have maintained thyroid stimulating hormone (TSH) levels <0.01 and thyroglobulin <0.20 and he have been asymptomatic until now.

DISCUSSION

It is estimated that the thyroglossal duct persist in 4% to 7% of the population in both genders and predominantly up to 15 years of age. The thyroglossal cyst carcinoma is rare, that is, less than 1% of cases, and it is predominantly in female young adults. Indications of malignancy such as pain, rapid growth, hoarseness, weight loss, and cervical lymph node enlargement obtained in the history must be carefully analyzed.[9]

About 80% of cases are found to be papillary carcinoma. And there is no consensus as to the etiology, being discussed the origin as metastases of a primary hidden thyroid tumor and other authors support the primary origin of thyroglossal cyst carcinoma.[9]

We must be aware that the origin of a thyroglossal cyst carcinoma lies in the fact that in about 62% of cases there are foci of thyroid tissue.[9] In our case report, we did not observe the criteria of Joseph and Komorowski (1975)[5] for the carcinoma of duct thyroglossal because we did not find foci of normal thyroid follicles in the cystic wall, and we did not have the presence of a normal thyroid gland.

Thus, as we did not identify the criteria of Kristensen et al. (1984),[10] in our study, as the surgical findings too, did not observe histologically normal ectopic thyroid follicles in the cyst, the tumor did not extend through the cystic wall, the thyroid gland normal and without involvement of cervical lymph nodes.

For the cases non operability according to Wexler (1996),[11] the conservative approach instead of total thyroidectomy should be emphasized, since a significant clinical percentage of microscopic findings of papillary thyroid carcinoma are questionable, since the cure rate reaches 95% of cases, which according to this author, its resection is not justified, which we disagree because we find cases that evolution for die to the clinical and anatomopathological findings similar to the find of our case related.

In our case report, we considered total thyroidectomy based on the criteria of Boswell et al. (1994),[12] who according to the author, these injuries cause clinical and loco-regional changes, and also images of changes in the duct of cyst that also caused greater potential for lymph and multifocal dissemination, being that the surgery has less common complications in the experienced hands, with a good prognosis of papillary carcinoma of thyroglossal cyst, meantime with reports of metastasis at a distance less than or equal to 2% of cases.

In this case report, as recommended by other author,[13] when performing the Sistrunk (1920)[8] operative technique, we observed the presence of the tumor in the thyroglossal cyst, whose anatomopathological exam showed to be papillary carcinoma and classic pattern, measuring about 1.8cm, grade 2, non-encapsulated, originating in thyroglossal cyst and infiltrating connective tissue, with a mitotic index of 2-4, vascular and lymphatic invasion not detected, peri-neural infiltration not detected and necrosis present in about 10%, with hyoid bone free of neoplasia.

Due to the presence of invasion of the thyroglossal cyst wall, without normal thyroid follicles in the duct, and with thyroid alterations, we indicate total thyroidectomy based on the literature, which found about 33% of the thyroid affected at the time of the definitive diagnosis of thyroglossal cyst carcinoma, as confirmed in this relate, the performance of total thyroidectomy based on the authors Almeida et al. (2012),[13] according to them, the presence of thyroid nodules in the evaluation of radiological exams and intraoperative findings of the tumor authorizes us to perform total thyroidectomy. We also followed the criteria of Peretz et al. (2004),[14] which indicate total thyroidectomy and should be recommended and followed by suppression of thyroid hormone and radioiodine therapy, which we performed in our case report.

In our case report, according to these authors,[9] the low-risk group indicates the surgical technique of Sistrunk (1920)[8] accompanied by hormonal suppression and case follow-up, however, according to Carter et al. (2014),[9] the characteristics of low-risk cases correspond to patients aged 15 to 45 years, a tumor in the thyroglossal duct cyst smaller than 4.0cm, without distant lymph node metastases, indicate the Sistrunk (1920)[8] operative technique without total thyroidectomy and without hormonal suppression and with follow-up of the case.

What in our case report, although we found a 42-year-old male patient, with evolution of a thyroglossal cyst for 12 months and progressive growth and an intraoperative tumor finding measuring 1.8cm and without suspicious cervical lymph node, we indicate total thyroidectomy despite the preoperative evaluation, the presence of thyroid with nodules in the right and left lobe with negative aspiration cytology for neoplasia.

In our study we agree with Katz and Hachigian (1988),[6] who consider that it is a metastasis of a hidden thyroid carcinoma and should be treated with total thyroidectomy and radioiodine therapy, in association to the Sistrunk (1920)[8] operative technique. Unlike Katz and Hachigian (1988)[6]

findings, most authors accept the proposed treatment, however, this theory[12] is based on the development of primary cancer in the thyroglossal cyst wall of an ectopic tissue, according to this author, in about 60% of the surgical specimens and the most of these cancers are papillary and generally cancer is limited to the cyst wall, but an invasion of adjacent tissue occurs 20.8% of cases[12] and lymph node metastasis in 8% to 11.5% of cases. According to Weiss and Orlich (1991),[15] they report that distant metastases (liver or lungs) due to papillary carcinoma of the thyroglossal cyst is about 1.8 % of cases.

Contrasting with the findings of Boswell et al. (1994),[12] when faced with thyroid cancer developed on the wall of the thyroglossal cyst, we have a question: is it a primary cancer of the thyroglossal cyst wall or is it a metastasis of thyroid cancer? The thyroglossal cyst wall may contain stratified or cubic epithelial lining of salivary gland tissue, from the gastrointestinal or thyroid tract, from which cancer may develop.[2] Papillary thyroid carcinoma is present in 80% to 90% of cases, followed by mixed carcinoma (follicular and papillary) and squamous cell carcinoma.[3] Pure follicular carcinoma and anaplastic carcinoma are extremely rare, as well as medullary carcinoma that has never been described. The cases of adenocarcinoma have not been well specified and the etiopathogenesis of this cancer remains unclear.[7]

Regarding the prognosis of cases of papillary carcinoma of the thyroglossal cyst submitted to the surgical technique of Sistrunk (1920)[8] it is generally curative based on the review by Weiss and Orlich (1991)[12] with 35 patients, who found four cases that also had thyroid gland carcinoma. Which makes us think that in about 80% of cases the papillary carcinoma of the thyroglossal cyst is primary in this region in the absence of alteration of the thyroid gland; however, since we found papillary thyroid carcinoma in thyroglossal cyst, we recommend total thyroidectomy in the presence of thyroid gland alterations, and we will thus be able to assess foci of papillary thyroid carcinoma, confirming the need for a greater number of cases to be established a relationship of metastasis in papillary carcinoma of the thyroglossal cyst.

The most authors indicate the total thyroidectomy and/or neck dissection in the presence of thyroid nodule or compromised lymph nodes found on clinical examination, complementary exams or in the trans-operative period without alterations in the thyroid gland and without suspicious palpable lymph nodes, both in physical examination and ultrasound and computed tomography exams of the neck. Which makes us think that in 80% to 88.6% of cases the carcinoma is primary in the cyst wall, therefore, most authors, with whom we agree, total thyroidectomy and/or neck dissection are only indicated in case of thyroid nodule or compromised lymph nodes found on clinical examination, complementary exams or in trans and postoperative. Weiss and Orlich (1991) [12] in a study with 35 patients, using the surgical technique of Sistrunk (1920),[8] reported that it was curative and found four cases that also had thyroid gland carcinoma.

In our study with the indication of total thyroidectomy and the finding of right lobe papillary microcarcinoma (0.2cm) and adenomatous goiter with unchanged thyroid hormones and antibodies, we indicated the suppression of thyroid-stimulating hormone (TSH), even with hormonal parameters within normal range. Radioiodine therapy with a total iodine dose of 131mCi and joint follow-up with the endocrinologist is also indicated. Regarding prognosis, we did not observe locoregional recurrence after surgery,[1] which most authors did not observe.[3]

Finally, in our case report, head and neck surgery had quarterly follow-up in the first year, four-monthly in the second year and semiannual in the third year and continuing the follow-up with the endocrinologist. For the treatment of recurrences or metastases, the possibilities of surgery, radiotherapy, or iodine therapy should be considered depending on the case.[2]

CONCLUSION

Even knowing that the development of the thyroglossal cyst is a rare event and its clinical presentation is like those with benign cyst, however, a rapid increase in cyst volume may suggest malignancy, and the diagnosis is rarely made before surgery and has confirmed with anatomopathological exam of the specimen. The surgery of choice, usually curative, is the operative technique of Sistrunk (1920).[8] Total thyroidectomy and neck dissection are indicated in cases of thyroid nodules and/or lymph nodes compromised by neoplasia.

Thus, considering the presence of papillary carcinoma of thyroglossal cyst, as a metastasis of microcarcinoma of thyroid cancer, it is based on the criteria of we do not finding normal thyroid follicles tissue foci in the thyroglossal cyst wall and in the presence of the thyroid alteration. The indication of hormone suppression, with hormonal parameters within hormonal range, as well as radioiodine therapy, were indicated by pathological finding prognostic and demographic factors. The prognosis is favorable, and follow-up was made in the first quarterly year, in the second year was made each four months, and the third year every six months with the respective TSH, thyroglobulin tests, and the neck ultrasound and chest radiography were annual with joint follow-up with the endocrinologist.

REFERENCES

1. Walton BR, Kock KE. Presentation and management of a thyroglossal duct cyst with a papillary carcinoma. South Med J. 1997 Jul;90(7):768-61.
2. Daour R, Aftimos G, Rahayel L. Papillary carcinoma arising in the wall of a thyroglossal cyst (in French). Ann Chir. 1996;50(2):162-4.

3. Sahraoui S, Zamiati S, Essadi M, Jalali K, Acharki UM, Ouhtatou F, et al. Papillary carcinoma arising from the thyroglossal duct remnants: review of literature and report a case. Rev Laryngol Otol Rhinol. 2000;121(3):161-3.
4. Bhagavan BS, Rao DR, Weinberg T. Carcinoma of thyroglossal duct cyst: case report and review of the literature. Surgery. 1970 Feb;67(2):281-92.
5. Joseph TJ, Komorowski RA. Thyroglossal duct carcinoma. Hum Pathol. 1975 Nov;6(6):717-29.
6. Katz AD, Hachigian M. Thyroglossal duct cyst. A third-year experience with emphasis on occurrence in older patients. Am J Surg. 1988 Jun;155(6):741-4.
7. Hilger AW, Thompson SD, Smallman LA, Watkinson JC. Papillary carcinoma arising in a thyroglossal duct cyst: a case report and literature review. J Laringol Otol. 1995 Nov;109(11):1124-7.
8. Sistrunk WE. The surgical treatment of cysts of the thyroglossal tract. Ann Surg. 1920 Feb;71(2):121-2.
9. Carter Y, Yeutter N, Mazeh H. Thyroglossal duct remnant carcinoma: beyond the sistrunk procedure. Surg Oncol. 2014 Sep;23(3):161-6.
10. Kristensen S, Juul A, Moesner J. Thyroglossal cyst carcinoma. J Laryngol Otol. 1984 Dec;98(12):1277-80.
11. Wexler MJ. Surgical management of thyroglossal duct carcinoma: is an aggressive approach justified?. Can J Surg. 1996 Aug;39(4):263-4.
12. Boswell WC, Zoller M, Williams JS, Lord SA, Check W. Thyroglossal duct carcinoma. Am Surg. 1994 Sep;60(9):650-5.
13. Almeida, et al. Carcinoma papilífero de cisto de ducto tireoglosso. Rev Bras Cir Cabeça Pescoço. 2012;41(2):96-8.
14. Peretz A, Leiberman E, Kapelushnik J, Hershkovitz E. Thyroglossal duct carcinoma in children: case presentation and review of the literature. Thyroid. 2004 Sep;14(9):777-85.
15. Weiss SD, Orlich CC. Papillary carcinoma of the thyroglossal duct cyst. Br J Surg. 1991 Jan;78(1):87-9.