

# Surgical treatment for papillary cyst carcinoma thyroglossus

## Abstract

**Introduction:** The thyroglossal cyst contains remnants of thyroid tissue that may give rise to a malignant neoplasm such as papillary carcinoma, which has been described in less than 1% of cases. This tissue originates at the base of the tongue in the foramen cecum, extending towards the anterior part of the neck.

**Materials and methods:** A retrospective and descriptive study was conducted, including all patients diagnosed with a thyroglossal cyst and treated at the Oncology Institute and Hospital Bernardo Del Valle S. INCAN from January 2010 to December 2019. The study involved the analysis of epidemiological data, diagnostic methods, and surgical treatments.

**Results:** The mean age at the time of diagnosis was 49.3 years. The most commonly requested study was ultrasound, conducted on 5 patients. When performing the fine-needle aspiration biopsy (FNAB), the Bethesda I result was common in 4 patients. The Sistrunk procedure was performed on 5 patients, and in 3 of those cases, an intraoperative frozen section study was not requested. Additionally, papillary carcinoma was reported in 1 patient, and in the final pathology, 4 cases of papillary carcinoma were diagnosed. Three patients underwent reoperation, resulting in a diagnosis of micropapillary carcinoma in one case and papillary carcinoma in another. The incidence at our institution was 57.1%.

**Conclusion:** The approach to surgical management is based on the pathology report of the Sistrunk surgery, taking into account the associated risk factors for each patient. Total thyroidectomy is considered when there is a risk of malignancy, and this approach contributes to effective treatment. Sharing the therapeutic experiences applied in these cases contributes to offering optimal treatment options.

**Keywords:** thyroglossal cyst, papillary carcinoma, sistrunk operation

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**Abbreviations:** FNAB, fine-needle aspiration biopsy; TSH, thyroid-stimulating hormone; CT, computerized axial tomography

## Introduction

The persistence of the thyroglossus conduit is a congenital anomaly resulting from thyroid gland migration during the embryonic period, occurring between the fifth and tenth week of gestation. This conduit begins at the base of the tongue in the foramen cecum and extends towards the anterior neck part. The persistence of the thyroglossal conduit occurs in approximately 7% of cases and may result in the accumulation of mucus or mucin, leading to a cyst. The thyroglossal cyst contains residual thyroid tissue that may give rise to malignancy, such as papillary carcinoma, which has been diagnosed in less than 1% of cases.<sup>1,2</sup> Papillary carcinoma in a thyroglossal cyst originates from ectopic thyroid tissue. Women are the most affected, with the average age of diagnosis being around 40 years old. It initially presents as a neck malignancy without symptoms, but gradually, symptoms will increase with time.<sup>3</sup> Surgical treatment of this pathology is not standardized. The initial surgery is based on the Sistrunk operation, which involves the resection of the thyroglossal cyst and the hyoid bone, thereby preventing the recurrence of these cases. In cases with a risk of malignancy, a total thyroidectomy is added to this procedure.<sup>4</sup>

## Materials and methods

A retrospective and descriptive study was conducted that included all patients diagnosed with a thyroglossal cyst who were treated at Bernardo Del Valle S. INCAN Cancer Institute and Hospital from January 2010 to December 2019. The study involved the analysis of epidemiological data, diagnostic methods, and surgical treatment.

## Results

Table 1 represents characteristics of patients treated for persistent thyroglossal cysts. The gender distribution showed 6 women and 1 man, with an average age of 49.4 years. The most frequently used imaging study was neck ultrasound in 5 patients, followed by computerized axial tomography in 2 patients. When diagnostic cytology was performed with a fine-needle aspiration biopsy (FNAB), 4 patients were classified as Bethesda I, while the remaining 3 patients received Bethesda II, III, and IV ratings. The Sistrunk surgery was performed in 5 patients, and in 2 patients, a Sistrunk surgery was combined with a total thyroidectomy. Intraoperative frozen section analysis (ETO) was not performed in 3 cases. Among the patients who underwent this analysis, two received negative reports; one of these patients was later diagnosed with papillary carcinoma, and the other one with follicular hyperplasia.

The post-surgical pathology results confirmed papillary carcinoma in 4 patients, while 2 were found to have thyroglossal cysts, and 1 had follicular hyperplasia. Based on these results, surgical reoperation was performed for patients diagnosed with malignancy to complete their treatment with a total thyroidectomy, which was carried out in 3 patients. The final pathology report for patients who completed surgical treatment indicated a case of micropapillary carcinoma, a case of papillary carcinoma, a case of thyroiditis, and 4 cases negative for malignancy. In our experience, treatment is primarily based on surgery, and follow-up includes the quantification of TSH (thyroid-stimulating hormone) and thyroglobulin in conjunction with neck ultrasound. In cases of papillary carcinoma in the thyroid gland, treatment is based on international guidelines for differentiated thyroid cancer.

**Table 1** Represents characteristics of patients treated for persistent thyroglossal cysts

Variable		Frequency	%
Age	32	1	14
	37	1	14
	41	1	14
	52	1	14
	57	1	14
	63	1	14
	64	1	14
Sex	Female	6	86
	Male	1	14
Image study	CT	2	29
	USG	5	71
Biopsy by needle aspiration fine (BAAF)	Bethesda 1	4	57
	Bethesda 2	1	14
	Bethesda 3	1	14
	Bethesda 4	1	14
Type of surgery	Sistrunk	5	71
	Sistrunk + Total Thyroidectomy	2	29
Intraoperative study	Papillary	1	14
	Follicular hyperplasia	1	14
	Negative	2	29
	Unrealized	3	43
Histology	Papillary carcinoma	4	57
	Follicular hyperplasia	1	14
	Thyroid cyst	2	29
Reoperation	No	4	57
	Total Thyroidectomy	3	43
Final pathology	Micropapillary	1	14
	Papillary	1	14
	No	4	57
	Thyroiditis	1	14
Total		7	100

## Discussion

Type of papillary carcinoma histology in cyst thyroglossus represents 78–85% of the cases, with its primary location being the cervical midline in 75% of cases. Only 10–25% can be located sideways in areas such as the submental, hyoid, thyrohyoid membrane, lingual, and suprasternal regions.<sup>5</sup> The main symptomatology involves the presentation of a small, smooth, and round nodule, sometimes accompanied by pain, redness, and signs of infection. To make this diagnosis, the main imaging studies requested are ultrasound and computerized axial tomography (CT). A sample for needle aspiration biopsy (FNAB) is also taken to obtain a cytological diagnosis.<sup>6</sup>

Thyroglossal cyst predominantly affects females, with women accounting for 86% of patients. The mean age was 49.4 years, which represents an older average age compared to the global statistics.<sup>3,7</sup> Despite its low incidence, a thyroglossal cyst may raise high suspicion of malignancy. Some guidelines recommend an intraoperative study (ETO) with the purpose of performing oncological surgery. Among the

surgical procedures described are the Sistrunk surgery and Sistrunk surgery plus total thyroidectomy, as there is a 11–25% probability of finding foci of papillary carcinoma in the thyroid. In exceptional cases, lymph node dissection is performed when there is evidence of disease.<sup>8,9</sup>

Although papillary carcinoma is a rare pathology, making the diagnosis can be challenging. There is no consensus for its treatment due to the limited number of cases reported in the literature. The recommended approach is to choose between the Sistrunk operation or Sistrunk operation plus total thyroidectomy, primarily if there are poor prognostic factors such as tumor size (greater than 1 cm) and tumor invasion of the wall.<sup>8,10</sup> The prognosis for papillary carcinoma is good, with a 95% survival rate at 10 years. The decision to perform a complementary thyroidectomy is based on the finding of malignancy in this study, with an individualized approach to treatment considering the risk-benefit ratio.<sup>9-11</sup>

Thyroglossal duct carcinoma had an incidence of 57.1% (4 cases) in the population studied over 9 years, which is higher than what is reported in the worldwide literature. This high incidence could be attributed to the fact that our institution is a national and Central American reference cancer center. It is possible that the malignancy of the thyroglossal cyst is related to the older age of the diagnosed patients.

## Conclusion

The diagnosis and treatment of thyroglossal duct carcinoma present challenges due to its low incidence. Surgical management is based on the pathology report, taking into account the individual risk factors of each patient. The performance of a total thyroidectomy in cases of malignancy contributes to a positive prognosis. Understanding the therapeutic experiences applied in adult patients enhances the overall management and treatment options.

One of the limitations of the study is the low incidence of reported cases and the little experience with its treatment.

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## Conflicts of interest

The authors, Dr. Maria Augusta Velasco and Dr. Juan Pazmiño, declare that there is no financial, real, or perceived conflict of interest in the publication of this manuscript.

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