



DOI: 10.14744/SEMB.2024.99390

Med Bull Sisli Etfal Hosp 2024;58(3):291–297

Original Research

Coexistence of Thyroglossal Cyst and Thyroid Disease in Adults: Surgical Outcomes From A Single Center

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Abstract

Objectives: Thyroglossal cysts (TGCs) usually present during childhood and before the age of 30, however, they can also be seen in adults, even in advanced age. Nodular thyroid disease is also common in adults. In the literature, there is an ongoing debate regarding the differences in clinical presentation, gender, and postoperative recurrence of TGC between children and adults. In this study, we aimed to process the data of adult patients who underwent surgery for TGC in our clinic, along with the data on concurrent thyroid disease and thyroid surgery.

Methods: The data of patients over 18 years old who were operated on for TGC at the General Surgery Clinic of Sisli Hamidiye Etfal Training and Research Hospital between 2018 and 2024 were retrospectively evaluated.

Results: A total of 16 patients with a mean age of 43.94 ± 12.98 (21–67) years, were included in the study (11 F/5 M). The diagnosis of TGC was made in 12 patients (75%) by ultrasonography (USG), in 1 patient (6.25%) by computed tomography, in 1 patient (6.25%) by magnetic resonance imaging (MRI), and in 2 patients (12.5%) incidentally intraoperatively. 13 patients (81.25%) underwent the Sistrunk procedure, and 3 patients (18.75%) underwent cyst excision. Among the 16 TGC patients, papillary thyroid cancer in the cyst was detected in one patient (6.25%) preoperatively. During preoperative evaluation, nodular thyroid disease was found in 12 patients (75%). Of these, papillary thyroid cancer was detected in 3 patients (18.75%) preoperatively. Of the TGC group, 3 (18.75%) underwent thyroidectomy for thyroid malignancy, and five (31.25%) underwent additional thyroid surgery for nodular thyroid disease. The patients were followed for a mean of 22.63 ± 18.32 months (3–67 months), and no recurrence of TGC was observed during the follow-up period.

Conclusion: In patients with TGC, thyroid diseases and the requirement for thyroidectomy due to benign or malignant thyroid disease are not uncommon. Patients with TGC should be evaluated for thyroid disease before surgical treatment. While the Sistrunk procedure is the standard surgical technique in the treatment of TGC, in adults, if the cyst terminates below the hyoid bone, total cyst excision without removing the central portion of the hyoid bone may be sufficient.

Keywords: Sistrunk procedure, thyroglossal cyst, thyroid disease

Please cite this article as "Yanar C, Cetinoglu I, Sengul Z, Caliskan O, Unlu MT, Aygun N, et al. Coexistence of Thyroglossal Cyst and Thyroid Disease in Adults: Surgical Outcomes From A Single Center. Med Bull Sisli Etfal Hosp 2024;58(3):291–297".

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Submitted Date: September 14, 2024 **Accepted Date:** September 28, 2024 **Available Online Date:** September 30, 2024

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Thyroglossal cysts (TGC) are congenital malformations that develop due to the failure of obliteration and persistence of the thyroglossal duct at any point along the migration path of the thyroid gland, which originates embryologically from a diverticulum at the base of the tongue.^[1]

The most commonly seen congenital neck lesions are TGCs and they account for approximately 70% of such cases.^[2]

Although TGCs develop in about 7% of the population, only a small part becomes symptomatic.^[3,4]

About 40% of TGCs present within the first 10 years of life, while 15% present after the age of 50.^[5]

Thyroid nodules are a common pathology, with a prevalence of approximately 70% when detected through high-resolution ultrasonography (USG).^[6] Although most do not require treatment, they should be evaluated for functionality, malignancy, and compression on surrounding structures.

In the literature, there is still an ongoing debate regarding the differences in the clinical presentation, gender, and postoperative recurrence of TGC between children and adults.^[3,7,8]

Additionally, to our knowledge, there is no study evaluating the frequency of thyroid diseases in these patients.

In this study, we aimed to assess the data on thyroid disease, thyroid surgery, and TGC in adult patients who underwent surgery for TGC in our clinic, in light of the current literature.

Methods

After obtaining approval from the Sisli Hamidiye Etfal Training and Research Hospital Ethics Committee (date: 03.09.2024, number: 4529), the data of patients over 18 years of age, who were operated on for TGC at the General Surgery Department of Sisli Hamidiye Etfal Training and Research Hospital between 2018 and 2024 were evaluated retrospectively. Patients whose preoperative, intraoperative, and postoperative follow-up data were unavailable were excluded from the study. The patients' preoperative clinical presentation, preoperative imaging methods, surgical techniques, the presence or absence of thyroidec-tomy, TGC localization, postoperative complications and recurrent disease, and follow-up duration (months) were assessed.^[9]

TGC was classified based on its vertical position as intral-in-gual, thyrohyoid, or suprasternal.^[9]

Recurrence was defined as the reformation of a cyst or fis-tula in the midline requiring surgical resection.^[10]

Results

A total of 16 patients meeting the study criteria were in-cluded, consisting of 11 females (68.7%) and 5 males (31.3%). The mean age of the patients was 43.94 ± 12.98 years (21-67 years). The characteristics of the patients are summarized in Table 1.

Table 1. The data of patients with TGC

	n
Age Mean+SD (min-max)	43.94+12.98 (21-67)
Gender (F/M)	11 (68.7%)/ 5(31.3%)
Preoperative complaint	
Swelling in the neck	15 (93.8%)
Incidental	2 (12.5%)
Preoperative evaluation	
Ultrasonography	16 (100%)
CT	2 (12.5%)
MRI	3 (18.75%)
Thyroid Scintigraphy	6 (37.5%)
Thyroid Function Tests	16 (100%)
FNAB	7 (43.75%)
TGC Diameter in the Imaging	28.9 mm (8-40)
TGC Presentation	
Primary	15 (93.8%)
Recurrence	1 (6.2%)
Preoperative Thyroid Disease	12 (75%)
Thyroid Malignancy	3 (18.75%)
Nodular Goiter	9 (56.25%)
Surgery Performed	
TGC Surgery	
Sistrunk Procedure	
TGC Excision	
Thyroid Surgery	8 (50%)
TT + BCND + SLND	2 (12.5%)
TT	5 (31.25%)
Lobectomy	1 (6.2%)
Pathology	
TGC Pathology	
Thyroid Tissue in the Cyst	10 (62.5%)
Papillary Thyroid Cancer	1 (6.2%)
Thyroid Pathology	
Papillary Cancer	3 (18.75%)
Nodular Hyperplasia	5 (31.25%)
Complication	
Tracheal Perforation and Mediastinitis	1 (6.2%)
Seroma	3 (18.75%)
Follow-up Duration	22.63+18.32 month
Mean+SD (min-max)	(3-67)
Recurrence	0

TT: Total Thyroidectomy; BCND: Bilateral Central Neck Dissection; SLND: Selective Lateral Neck Dissection.

Preoperative Symptoms

15 patients (93.8%) presented with swelling in the neck due to TGC or thyroid disease, while 1 patient was diagnosed during a general examination. None of the patients had infection, abscess, or discharge due to a fistula from the neck related to TGC.

Previous Surgeries

4 patients had undergone prior surgeries for thyroid or TGC at external centers. One patient had total thyroidectomy for papillary thyroid cancer 17 years ago, a subsequent operation for local recurrence 14 years ago, lateral neck dissection 12 years ago, and TGC excision 1.5 years ago. The other three patients had undergone subtotal thyroidectomy in the time period of 17-30 years ago. One of these patients had persistent unilateral left vocal cord paralysis and permanent hypoparathyroidism.

Preoperative Imaging

All patients underwent preoperative USG. USG revealed a TGC-compatible appearance in 12 patients (75%). In the recurrent TGC case, the lesion was visualized as two separate cystic lesions. Additionally, nodular thyroid disease was detected in 12 patients (75%).

Thyroid Scintigraphy

6 patients (37.5%) underwent thyroid scintigraphy with Tc99m pertechnetate. Activity uptake was detected in the TGC region in one patient (16.7%).

Cross-Sectional Imaging

Computed tomography (CT) was performed on two patients. One patient was diagnosed with TGC by CT (6.25%). The other patient, who was initially identified by USG and suggested for further examination, was confirmed with TGC by CT. MRI was conducted for 3 patients. One patient was diagnosed by MRI (6.25%) and the other two patients were confirmed with the diagnosis of TGC, who were previously examined by USG and recommended further examination.

In two patients, preoperative imaging did not identify TGC. One of these patients had a large multinodular goiter with compressive symptoms, and TGC was found intraoperatively. The other patient, diagnosed with papillary thyroid cancer, had TGC detected and excised during central neck dissection.

Fine Needle Aspiration Biopsy (FNAB)

8 of these patients also underwent thyroidectomy. FNAB was performed on a total of 7 patients (43.75%). Of these, 2 patients had FNAB of both thyroid nodules and cervical

lymph nodes. Malignancy (Bethesda 6: compatible with papillary thyroid cancer) was found in both the thyroid nodules and cervical lymph nodes. Among the patients who had FNAB only on thyroid nodules, one was malignant (Bethesda 6: compatible with papillary thyroid cancer) and three were benign (Bethesda 2). One patient had FNAB of a nodular structure within the TGC, revealing malignancy (Bethesda 6: compatible with papillary thyroid cancer).

Cyst Size on Imaging

The largest diameter of the cystic lesion ranged from 8 to 40 mm on USG, CT, and MRI.

Surgical Intervention

Thirteen patients (81.3%) underwent Sistrunk surgery due to TGC, while three patients (18.7%) underwent cyst excision.

In the Sistrunk procedure, the TGC remnant is removed along with the central portion of the hyoid bone up to the base of the tongue.^[11]

In patients who underwent cyst excision, the central portion of the hyoid bone was not removed. In one of these patients, the thyroglossal cyst tract extended anteriorly from the hyoid bone, so the tract was removed without removing the hyoid bone. In the other two patients, the thyroglossal cyst tissue terminated inferior to the hyoid bone, so superior dissection was not performed.

Cyst Localization

TGCs were located in the suprahyoid region in 1 patient (6.25%) and thyrohyoid region in 15 patients (93.75%).

Additional Surgical Procedures

In two patients with preoperative diagnosis of papillary thyroid cancer and lateral metastasis, total thyroidectomy, bilateral central neck dissection, and selective lateral neck dissection were performed. Five patients underwent total thyroidectomy, and one patient underwent lobectomy. There was no indication for thyroid intervention in four patients with nodular thyroid disease.

Pathology

Of the 16 TGCs removed, thyroid tissue was found in 10 patients (62.5%). Papillary thyroid cancer (classical type, 2.5 cm in diameter, without lymphovascular invasion) was identified in the thyroid tissue of one patient (6.25%). Multicystic ducts were described in the pathology of one patient.

In patients who underwent thyroidectomy:

- One patient had multifocal diffuse sclerosing-type papillary thyroid carcinoma with central and lateral metastases.

- One patient had tall cell-type papillary thyroid carcinoma with central and lateral metastases.
- One patient had papillary thyroid carcinoma limited to the thyroid.

In the patient with papillary thyroid carcinoma associated with TGC, nodular hyperplasia was found in the thyroid tissue, and nodular hyperplasia was also observed in the thyroid glands of the other four patients who underwent thyroidectomy.

Postoperative Complications

Seroma developed in the TGC region in three patients but resolved without surgical intervention. One patient experienced a major complication that required surgical intervention, which was not related to TGC. This patient had a perforation of the trachea and developed mediastinitis on postoperative day 10 following thyroidectomy. The patient recovered with treatment but unfortunately died in the 5th year due to esophageal cancer.

Radioactive Iodine Therapy (RAI)

RAI was administered postoperatively to three patients with TGC, including two with lateral metastases and one with cancer in the cyst.

Follow-Up

The mean follow-up period for patients was 22.63 ± 18.32 months (ranging, 3-67 months). During this time, no recurrence of TGC was detected. Additionally, no recurrence of malignancy was observed in patients with malignant cases.

Discussion

Thyroglossal cysts are typically observed in childhood and usually present before the age of 30, but they can also occur in adults and even in older age.^[3,12,13]

Our study focused on patients over the age of 18, with a mean age of approximately 44 years. A meta-analysis including 1,316 patients stated that 34.6% of patients were over 30 years old.^[14]

Similarly, a study evaluating adult patient outcomes from the American College of Surgeons National Surgical Quality Improvement Program (NSQIP) database between 2005 and 2014 reported a mean age of 44.3 years, which is comparable to our findings.^[8]

Although TGCs are embryologically observed equally in both genders, different studies report varying gender ratios.^[4]

In a review including approximately 1,750 patients, the gender distribution was found to be equal between males and females.^[9]

In our study, the proportion of female patients was higher, with approximately two-thirds of the patients. This may be related to the fact that our study focused on adult patients and that three-quarters of the patients had nodular thyroid disease. In another study evaluating adult patients, 57% were female and 43% were male.^[13]

In a study that included both pediatric and adult patients, a similar finding was reported, with the proportion of female patients being 58%.^[14]

However, there are also studies indicating that approximately 60% of patients are male, both in children and adults.^[10,12]

Despite the high prevalence of nodular thyroid disease, many nodules do not require further investigation or surgical intervention. Our center, a high-volume endocrine surgery center, found that 75% of patients with TGC had thyroid malignancy or nodular goiter, and half required thyroid surgery due to either malignant or benign thyroid disease. Except for one patient, all administered medical attention due to swelling in the neck related to TGC or thyroid disease. In two cases, TGC was incidentally discovered during thyroidectomy, despite the absence of preoperative findings related to TGC. In the literature, the most common reason for presentation is painless neck mass.^[5] Secondary manifestations of TGCs include cyst infections and thyroglossal fistulas. A recent meta-analysis evaluating clinical presentations and symptoms in 1,015 patients found the following prevalence rates: cervical cystic masses in approximately 75% (95% CI: 0.72-0.79), cervical infections or abscesses in about 34% (95% CI: 0.31-0.37), fistulas or draining sinuses in approximately 18% (95% CI: 0.15-0.22), dysphagia in 9% (95% CI: 0.07-0.11), and airway obstruction in about 6% (95% CI: 0.03-0.09).^[15]

In a study comparing children and adults, the most common presentation in adults was a painless mass in approximately 90% of cases, while in children, 53.5% of patients presented with a tracheal fistula.^[16]

In another study, the most common presentation in both children and adults was a neck mass, with thyroglossal fistula occurring in approximately 10% of cases. Thyroglossal fistulas were found to be twice as common in children compared to adults.^[17]

Ultrasound is the ideal initial imaging method for evaluating the solid or cystic nature of neck masses and assessing their relationship with surrounding structures.^[18]

At our center, USG is the preferred initial imaging method for evaluating thyroid and neck lesions, and all patients underwent USG. In this study, TGC was diagnosed by USG in 75% of patients, while one patient was diagnosed by CT

and one by MRI. Additionally, in two patients where the cyst was identified by USG, it was confirmed by CT in two cases and by MRI in another two cases. Of the six patients who underwent thyroid scintigraphy, one showed uptake in the cyst region.

When necessary, for evaluating cystic neck lesions in adults, CT or MRI can be used as complementary methods or for assessing recurrences, especially when USG does not provide sufficient imaging.^[18]

Although USG is generally the most commonly used imaging method for evaluating TGCs in both adults and children, some centers report that USG is the most frequently used test in children, while CT is more commonly used in adults.^[10,14]

Absence of uptake or minimal uptake in the cyst wall on thyroid scintigraphy does not significantly contribute to the diagnosis. However, scintigraphy is important, especially in children, to exclude the presence of ectopic median thyroid tissue when the mass shows a solid structure. Median ectopic thyroid tissue can be the patient's sole functional thyroid tissue.^[19]

TGCs can be localized anywhere along the embryological migration path of the thyroid gland, between the base of the tongue and the suprasternal notch.^[9] In our study, TGCs were localized as follows: 15 cases (93.8%) in the thyrohyoid region and 1 case (6.2%) in the suprahyoid region. The literature reports the localization distribution as follows: intralingual in 2.1%, suprahyoid in 24.1%, thyrohyoid in 60.9%, and suprasternal in 12.9%.^[9] The low incidence of suprasternal localization in our study may be related to the inclusion of adult patients and the evaluation of suprahyoid lesions by the otolaryngology department at our center.

Occasionally, malignancy can be found within TGC tissue. Clinical series report an incidence ranging from 1% to 7%, with more than 95% of these cases originating from the thyroid tissue.^[17,20-22]

Many carcinomas originating from thyroglossal cysts cannot be clinically differentiated from TGCs.^[9]

In this study, based on USG findings, FNAB was performed on one patient due to solid tissue in the TGC, and/or on thyroid and/or neck lymph nodes. Papillary thyroid carcinoma diagnoses were established preoperatively through FNAB of both the thyroid and TGC tissues.

Thomson et al.^[17] conducted a study, which involved 22 malignancies in TGCs with thyroid origin. In their study, preoperative FNAB was performed on 12 cases. The cytopathological results were classified as Bethesda IV (follicular neoplasm)-V (suspicious for malignancy)-VI (malignant)

in 6 cases, Bethesda II (benign) in 4 cases, and Bethesda I (nondiagnostic) in 2 cases.

In large series, the false-negative rate of FNAB for suspicious malignant tumors originating from thyroglossal cysts ranges between 40% and 60%.^[17,23,24]

In a study involving 82 TGCs with FNAB, only 2 out of 6 malignancies were detected. The sensitivity in this study was only 33%, which is insufficient for excluding malignancy. Therefore, for suspicious thyroglossal cysts, especially when considering papillary thyroid carcinoma, it is recommended to perform a frozen section to help exclude malignancy.^[25]

However, in extensive case series where clinical, radiological, and cytological features of TGCs were evaluated together, it was reported that combining clinical and radiological findings with cytological results of USG-guided FNAB, facilitated both the diagnosis and differential diagnosis of TGCs.^[26]

As expected from the embryological development of TGC, thyroid tissue may be found in the cyst wall. The incidence of ectopic thyroid tissue reported in the literature ranges between 26.5% and 71%.^[17,21,27] In our study, thyroid tissue was detected in 62.5% of the cyst walls.

Reviews published in the literature report a general recurrence rate of 7.3% to 11% following surgical treatment of TGC.^[10,15,28]

Several predisposing factors for recurrence have been defined. These include incomplete excision of the cyst, multiple tracts, multicystic lesions, children under the age of 3, preoperative infection, intraoperative cyst rupture, postoperative infection, and recurrent TGC.^[10,12,17,19,28,29]

The most significant factor affecting recurrence is incomplete surgical resection, and Sistrunk procedure is considered as the standard surgical treatment for TGC.^[17] Recent two large studies have shown that the recurrence rate is 3-5% for Sistrunk surgery, whereas it ranges from 55-70% for simple cystectomy.^[10,17]

The age of the patient has also been reported to be associated with recurrence. In a study comparing recurrence rates based on the age at which Sistrunk surgery was performed for TGC, the recurrence rates were found to be 38.9% in children under 3 years, 16.7% in children aged 3-18 years, and 3.6% in adults. The study indicated that the recurrence rate is significantly lower in adults compared to the pediatric age groups.^[12]

Rattan et al.^[16] evaluated data from 43 children and 28 adults who underwent surgery for TGC. In all children, the Sistrunk procedure was performed, while 21.43% of adults received the Sistrunk operation. The remaining 78.57% of

adults underwent cyst excision, where the upper end of the thyroglossal tract was removed without reaching the hyoid bone, due to the loss of tract firmness and the soft tissue spreading in a fan-like manner. Recurrence was observed in one patient in each group. The researchers suggested that this early termination of the tract, observed only in adults, might be attributed to long-term fibrosis of the thyroglossal tract with age. They indicated that if TGC in adults ends macroscopically without reaching the hyoid bone, it could be treated effectively with simple excision.^[16] More than %85 of the recurrences occur within the first year.^[10,12]

However, there is no definitive time frame established for excluding recurrences. Most experts agree that the majority of recurrent cases are diagnosed within a few weeks after the initial surgery.^[28]

In our study, no recurrences were observed in either the patients who underwent Sistrunk surgery or those who had only cyst excision, with an average follow-up period of 22.63 months. In one patient who had a cyst excision, the tract was removed without resecting the middle portion of the hyoid bone, as the tract extended anterior to the hyoid bone. In two other patients, similar to the findings of Rattan et al.,^[16] the TGC ended inferior to the hyoid bone. These findings suggest that for TGCs ending inferior to the hyoid bone, cyst excision alone might be sufficient. We believe that total cyst excision can be applied to TGCs that do not extend up to the level of the hyoid bone with careful dissection.

The main limitations of our study are its retrospective design and the limited number of cases. Additionally, this study may contribute to the literature due to its sufficient follow-up period and its evaluation of the prevalence of thyroid disease.

Conclusion

In conclusion, the need for thyroidectomy due to thyroid disease, whether malignant or benign, is not uncommon in patients with TGC. These patients should be evaluated for thyroid disease before surgical treatment. While Sistrunk procedure is the standard surgical technique for TGC, in adults, if the cyst ends inferior to the hyoid bone, total cyst excision without removing the middle portion of the hyoid bone may be sufficient.

Disclosures

Ethics Committee Approval: The study was approved by the Sisli Hamidiye Etfal Training and Research Hospital Ethics Committee (Number: 4529, Date: 03.09.2024).

Peer-review: Externally peer-reviewed.

Financial Support: No financial support was declared by the authors.

Patient Informed Consent: Written informed consent was obtained from the patients.

Conflict of Interest: No conflict of interest was declared by the authors.

Authorship Contributions: Concept – M.U., N.A., C.Y.; Design – M.U., N.A., C.Y.; Supervision – M.U., N.A., M.T.U.; Materials – M.T.U., O.C., C.Y., I.C., Z.S.; Data collection &/or processing – M.T.U., O.C., C.Y., I.C., Z.S.; Analysis and/or interpretation – M.U., N.A.; Literature search – M.U., N.A.; Writing – M.U., N.A., I.C.; Critical review – M.U., N.A., I.C.

Use of AI for Writing Assistance: None declared.

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