European Journal of Therapeutics

DOI: 10.5152/eurjther.2021.20119

Original Article

Thyroglossal Duct Cysts: A Clinico-Surgical Experience of 100 Cases

İsmail Aytaç, Orhan Tunç

Department of Otorhinolaryngology, Gaziantep University Faculty of Medicine, Gaziantep, Turkey

ABSTRACT

Objective: Thyroglossal duct cysts occur if a thyroglossal duct does not disappear after the complete embryonic development of the thyroid gland and becomes cystic. This study aimed to examine the clinical features, physical examination findings, and treatments of 100 patients who underwent surgery with the diagnosis of thyroglossal duct cyst in the midline of the neck.

Methods: This was a retrospective study based on anamnesis forms, clinical examinations, and radiographic imaging of 100 cases diagnosed with and operated on for thyroglossal duct cyst in our clinic.

Results: Of the 100 patients, 53 were males and 47 were females, with their ages ranging from 1 to 62 (mean 18.15 ± 15.8) years. The most common complaints were neck swelling (88%) and intermittent discharge (11%). Concomitant infection and fistula were present in 52 and 30% patients, respectively. The most common localization observed in 67 (67%) patients was in the infrahyoid area. Infection and abscess were observed in six cases (6%) during the early post-operative period. Papillary thyroid carcinoma, in addition to the cyst, was found in four cases. Recurrence was observed in three (3.7%) of the 81 primary cases. Sistrunk procedure was employed in all study patients.

Conclusion: Thyroglossal duct cyst is the most common observed congenital mass in the neck. It must be considered in the differential diagnosis of patients admitted due to discharge and swelling in the midline of the neck. The generally accepted treatment of thyroglossal duct cysts is surgery, with Sistrunk surgery being the most appropriate surgical technique owing to its low complication and recurrence rate. It should also be known that these cysts have a risk of malignant transformation. The most common post-operative complications observed in our study were infection and abscess.

Keywords: Thyroglossal, cysts, fistula, midline neck swelling, sistrunk

INTRODUCTION

The thyroglossal duct forms during the embryonic development of the thyroid gland. While the thyroid rudimentary base descends caudally in the neck, it is believed that the tongue forms a channel attached to the origin point at the foramen cecum level.[1,2] Thyroglossal duct typically undergoes atrophy during the 7-10th gestational week, following the primitive thyroid's transition to the last pretracheal position in the neck's lower half. Thyroglossal duct cysts occur if the thyroglossal duct does not disappear after the thyroid gland completes its embryonic development and becomes cystic.[3,4] This developmental abnormality is observed in approximately 7% of the population.[1,3,5] Thyroglossal duct cysts are typically present in the neck's midline and are painless swellings that move with tongue movement; they can also be fistulized to the skin, in which case, they are called thyroglossal fistula.[3]

Thyroglossal duct cysts, although not infrequent in adults,[6] are the most common cervical lesions encountered in infancy and childhood.[5] They constitute >75% of the midline neck masses in childhood.[7] Cysts can occur anywhere along the migration path of the thyroglossal duct of a developing thyroid gland. Histologically, it is covered by the respiratory epithelium, squamous epithelium, or a combination of both. Microscopic foci of ectopic thyroid tissue are variable.[8] Thyroglossal duct cyst is commonly observed between the hyoid and thyroid gland, but differences are present in the admission levels for the same. Approximately 20–25% are present at the suprahyoid level, 15–20% at the hyoid level, and 25–65% at the infrahyoid level.[3]

Cyst infection and accompanying fistula are common, especially during the first decade of life. If a fistula is present, the fistulized skin must be removed along with the cystic structure and hyoid bone corpus.[1]

Approximately 1% patients with thyroglossal duct cysts may have an associated malignancy, which may of different types (papillary, follicular, anaplastic thyroid cancer, Hurtle cell carcinomas, etc.).[9] However, the most common type of malignancy is papillary thyroid carcinoma.[4,10] Due to the risk of malignancy, the treatment of this condition is surgical.[11] Sistrunk operation is the gold standard in surgery for this condition, involving the removal of hyoid bone corpus together with the entire cyst tract.[12–14]

How to cite: Aytaç İ, Tunç O. Thyroglossal Duct Cysts: A Clinico-Surgical Experience of 100 Cases. Eur J Ther 2021; 27(2): 118-122.

ORCID iDs of the authors: İ.A. 0000-0002-0947-366X; T.O. 0000-0001-7764-1138.

Corresponding Author: İsmail Aytaç E-mail: dr.iaytac@gmail.com

Received: 15.12.2020 • **Accepted:** 18.02.2021

In this study, we examined the symptoms, findings, revision rates, post-operative complications, and follow-up of patients who underwent Sistrunk surgery with the diagnosis of thyroglossal cyst or fistula in our clinic.

MATERIAL AND METHODS

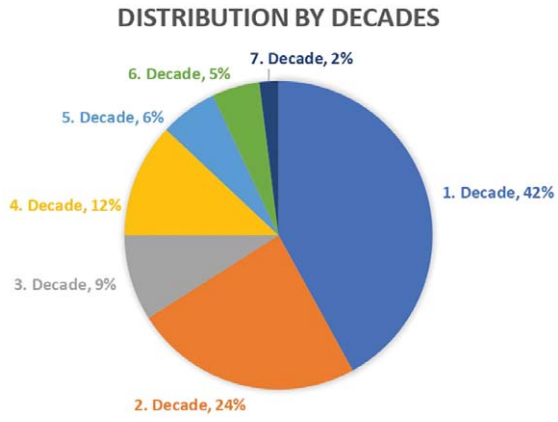
We included 100 patients who underwent surgery with the diagnosis of thyroglossal duct cyst in our hospital's clinic between 2003 and 2020. Patient profiles were examined for sex, age, admission complaints and findings, cyst location with respect to the hyoid bone and neck midline, concomitant infection, thyroglossal duct cyst fistulization, imaging findings, operation performed, post-operative recurrence and complications, post-operative pathology results, and patient follow-ups. Additionally, the obtained findings were evaluated with the existing literature. Descriptive statistical analysis, χ^2 and Crosstabs, was performed using Statistical Package for the Social Sciences (SPSS) version 22 (IBM SPSS Corp., Armonk, NY, USA). The statistical significance was set at $P < .05$. This study was conducted in accordance with the principles of the Declaration of Helsinki. Additionally, approval for this study was obtained from the Ethical Committee of the Faculty of Medicine of Gaziantep University (2020/329).

RESULTS

Of the 100 patients who underwent surgery, 53 (53.3%) were males and 47 (47%) were females. Their ages were 1-62 (mean 18.15 ± 15.837) years. Admission was most frequently observed in the first decade of life (46%) (Figure 1). The most common complaints at the time of first admission were swelling in the neck (88%) and discharge in the neck (11%), and more rarely, difficulty in swallowing (1%). Additionally, 30% patients had a fistula mouth in the neck or under the chin. Examination findings revealed that 30 patients had a fistula and 70 had a cyst. Additionally, 52% patients had an infection. Patients with cyst formation had a well-circumscribed, mobile, and painless swelling, while those with fistulization had a fistula mouth on the skin. Neck ultrasonography (USG) imaging records were available for all patients. USG findings revealed a cystic or fistulized cystic lesion with debris and dense contents. All patients underwent Sistrunk surgery under general anesthesia.

In the follow-up of 81 patients whose primary surgery was performed by us, recurrence was observed in only three patients (3.7%), and these patients were reoperated at the same incision site. Additionally, 19 patients previously had been operated in another center. The excision of the hyoid bone corpus was either not performed or was performed insufficiently in these patients. Hence, hyoid bone corpus excision and Sistrunk operation were performed in all these patients. As recurrence was observed in two patients operated in an external center and reoperated by us, these patients had to undergo surgery for a third time as well. No complications were observed later during their follow-ups.

Figure 1. Distribution of thyroglossal duct cysts in patients according to decades of life (100 patients).



Infection was observed in six of the 100 patients during the post-operative follow-up period (6%). Three of these patients required surgical drainage, while the other three were treated using antibiotherapy without drainage. Histopathological examination results were in accordance with thyroglossal duct cysts for all patients. The results of four patients revealed signs of malignancy (papillary thyroid carcinoma on the thyroglossal background) in addition to the cyst. Three of these patients underwent thyroidectomy in the same session, while one underwent thyroidectomy later. When the USG results were evaluated, the location of the cyst was at the infrahyoid level in 67 patients (67%), the suprahyoid level in 20 patients (20%), and the hyoid level in 13 patients (13%).

The most common complaints at admission were painless swelling in the neck midline (88%) and intermittent discharge (11%). Additionally, only one patient with a suprahyoid cyst had difficulty in swallowing. When the relationship between the localization of the cyst and the complaints at admission was evaluated, no significant relationship was found (Table 1).

The USG dimensions of thyroglossal duct cysts were 2-47 mm, the mean size being 19.38 ± 10.180 mm. When the relationship between the cyst's size and the recurrence rates was evaluated, no statistically significant relationship was found ($P > .05$).

Main Points

- Thyroglossal duct cyst is the most common congenital neck mass in the midline of the neck.
- Thyroglossal duct cysts are typically present in the neck's midline and are painless swellings that move with tongue movement.
- Cyst infection is a common condition often accompanied by the presence of a fistula.
- Malignant degeneration can occur; hence, it must be treated surgically.
- Sistrunk operation is the gold standard in surgery for this condition, involving the removal of hyoid bone corpus together with the entire cyst tract.

DISCUSSION

Thyroglossal duct cyst is the most common congenital neck mass in the midline of the neck, which is caused by dilatation

Table 1. Comparison of Cyst Localization and Admission Complaints

Admission complaint	Hyoid localization	Infrahyoid localization	Suprahyoid localization	Total
Discharge	2	6	4	12
Neck swelling	11	61	15	87
Difculty in swallowing	0	0	1	1
Total	13	67	20	100

and persistence of epithelial tract remnants in thyroid migration during embryogenesis.[4,14] These anomalies, generally observed as cysts, are called thyroglossal fistulas if they open to the neck epithelium.[7]

The exact incidence of thyroglossal canal remnants is unknown. However, it has been reported that a thyroglossal duct cyst or remnant is observed in about 7% of the population.[1,4,5]

Although these anomalies, observed equally in both sexes, can occur in any decade of life, they are more noticeable in the childhood, especially in the first 5 years of life.[6,15] In the present study, the age range was 1-62 years, and in correlation with the literature, 46% of the patients were in their first decade of life. Additionally, 53 (53%) patients were males and 47 (47%) were females.

According to research, approximately 50% of thyroglossal duct cysts become infected, which is the main reason for patients consulting a physician.[3,7,16] Consistent with the literature, infected cysts were observed in 52 patients (52%) in the present study. Cyst infection is a common clinical picture often accompanied by a fistula.[17] In a study conducted by Abuabara et al.,[18] it was reported that one-fourth of the adult patients presented with a drainage sinus caused by spontaneous drainage or surgical drainage of the abscess. Additionally, Ren et al.[15] reported cutaneous fistula in 10% patients. In this study, thyroglossal fistula was observed in 30 (30%) patients, with 28 of these having an active infection. In patients having a fistula, the fistulized skin was excised along with the cystic structure and the hyoid bone corpus (Figure 2). Consistent with the literature,[1,7,14] the fistula appeared during the first decade of life in half of the patients.

When the cyst is infected, pain, difficulty in swallowing, and skin hyperemia may be observed.[19] Thyroglossal cysts are rarely associated with dysphagia or airway obstruction.[20] Although the order of priority varies between studies, the most common complaints were painless swelling and intermittent discharge.[7] A comprehensive meta-analysis study performed on 1,015 cystic cervical masses reported infection, abscess, fistula, dysphagia, and airway obstruction to be the most common clinical presentations and symptoms.[11] The most common complaint in our patients was painless swelling in the neck midline (88%) and intermittent discharge (11%), with difficulty in swallowing being reported in only one case with a suprahyoid cyst. Due to its embryological origin, the thyroglossal duct cyst is usually present in the midline of the neck, in the form of a somewhat hard mass that moves as the tongue is pulled out.[4,11,21] These cysts can be found anywhere along the thyroglossal duct, from the base of the tongue to the suprasternal notch.[18] However, in approximately three-fourth of the cases, they are observed under the hyoid bone in the neck.[6,20,22] Based on the rates determined in previous studies, the most common locations of cysts are the infrahyoid region (61%), the suprahyoid region (24%), the suprasternal region (13%), and the intralingual region (2%).[5,9] In accordance with the literature, 67 patients (67%) in our study had infrahyoid cysts, 20 (20%) had suprahyoid cysts, and 13 (13%) had hyoid cysts. An intralingual cyst was not observed in any of the patients. The differential diagnosis should consider dermoid cyst, cystic hygroma, branchial cyst, thyroid pyramidal lobe hyperplasia and cysts, lipoma, ectopic thyroid tissue, lymphadenopathy, sebaceous cysts, hamartoma, hemangioma, teratoma, and primary or metastatic neoplasms.[5,7,11] USG, computed tomography (CT), and magnetic resonance imaging (MRI) are important auxiliary diagnostic methods for determining the cyst's size and its surrounding tissues. On USG, it is observed as homogeneous hypoechoic or heterogeneous lesions. On CT, it

Figure 2. Thyroglossal duct cyst incision line and inclusion of fistulized skin.

is observed as a hypodense mass and thickening of the wall. On MRI, it is more homogeneous in T1 than normal cysts and fluids.[23] Neck USG is a noninvasive and appropriate imaging method for diagnosing thyroglossal cysts.[11,18] However, USG may not reveal the depth of the hyoid and infrahyoid thyroglossal duct cysts and cannot reliably evaluate the base of the tongue in suprahyoid thyroglossal duct cysts. CT scan is better for understanding the relationship between the cyst and the hyoid. MRI is preferred for lesions close to the base of the tongue.[21] Thyroid scintigraphy can be performed to differentiate the presence of ectopic thyroid tissue.[24] The accepted treatment for thyroglossal duct cysts is surgery. Recurrent infections, unwanted cosmetic appearance, malignant degeneration, and fistula formation are surgical indications for thyroglossal duct cysts and fistulas.[11,25] Sistrunk surgery is frequently performed these days, which involves making a horizontal incision on the lower edge of the mass and carefully dissecting the cyst from the skin. After the cyst is freed, the cyst tract, along with the hyoid bone corpus, is dissected toward the tongue base. To prevent recurrence, dissection is continued up to the foremen cecum and the cyst is completely removed along with the hyoid corpus and the tract by placing a suture.[4,12–14] Post-operative recurrence rate after this procedure is 1.5–4%.[23] However, the recurrence rate has been reported to be 38% in cases where the hyoid bone is not resected.[19] If the hyoid corpus is not removed, this rate may increase up to 85%.[19,23,26] During surgery, care should be taken not to damage the vagus, hypoglossus, spinal accessory nerve, and other important structures adjacent to the tract.[19] Sistrunk surgery was performed in all of the study patients in our clinic. It has been reported that approximately 1% of patients with thyroglossal duct cysts present with malignancy, the most common being papillary thyroid carcinoma.[3,4,11,25] In this study, papillary thyroid carcinoma occurred four patients with thyroglossal duct cysts, and Sistrunk + thyroidectomy surgery was performed on these patients.

There are many reasons for unsuccessful surgical treatment of thyroglossal duct cyst. It has been demonstrated that an incorrect surgical procedure, such as a cystectomy alone, is the primary reason behind the high recurrence rate.[28] Sistrunk[12] suggested that various factors, such as the patient's young age, infection, and cyst's puncture during surgery, also caused recurrence. Ein et al.[29] examined 270 patients who underwent surgery for thyroglossal duct cysts and reported that infection and drainage of the cyst were the most important factors leading to recurrence. Dedivitis et al.[30] reported that the diagnosis of thyroglossal duct cyst could be made clinically, and that Sistrunk operation showed good results with low complication (11.62%) and recurrence (2.32%) rates; they also stated that antibiotic treatment could be avoided, and that hospital stay was short. Considering this information, thyroglossal duct cysts should also be considered in the differential diagnosis of patients presenting with neck swelling or mass. Additionally, it should not be forgotten that Sistrunk surgery is the gold standard in the treatment of this condition. Sistrunk surgery was performed for all cases in our clinic. The recurrence rate was 3.7% in patients whose primary surgery was performed by us, wherein these operations were performed by various surgeons as the clinic where the study was conducted is also an educational hospital. It is believed that the recurrence rate may be even lower if the surgeries are performed by the same surgeon.

CONCLUSION

Thyroglossal duct cyst is the most common congenital neck mass in the midline of the neck. Anamnesis and physical examination as well as radiological examinations are typically used for diagnosis. Cyst infection is a common condition often accompanied by the presence of a fistula. Although rare, malignant degeneration can occur; hence, it must be treated surgically. The most appropriate surgical technique is the Sistrunk surgery, which has fewer cases of recurrences.

Ethics Committee Approval: This study was approved by the Ethics Committee of the Faculty of Medicine of Gaziantep University (2020/329).

Informed Consent: N/A

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - I.A.; Design - I.A.; Supervision - O.T.; Resources - I.A. and O.T.; Materials - I.A. and O.T.; Data Collection and/or Processing - I.A. and O.T.; Analysis and/or Interpretation - I.A. and O.T.; Literature search - I.A.; Manuscript Writing - I.A.; Critical Review - O.T.; Other - I.A. and O.T.

Conflict of Interest: The authors have no conflicts of interest to declare.

Financial Disclosure: The authors declared that this study has received no financial support.

References

1. Thompson LDR, Herrera HB, Lau SK. A clinicopathologic series of 685 thyroglossal duct remnant cysts. *Head Neck Pathol*. 2016;10:465-474. [CrossRef]
2. Chou J, Walters A, Hage R, et al. Thyroglossal duct cysts: Anatomy, embryology and treatment. *Surg Radiol Anat*. 2013;35(10):875-881. [CrossRef]
3. Soni S, Poorey VK, Chouksey S. Thyroglossal duct cyst, variation in presentation, our experience. *Indian J Otolaryngol Head Neck Surg*. 2014;66(4):398-400. [CrossRef]
4. Narayana Moorthy S, Arcot R. Thyroglossal duct cyst more than just an embryological remnant. *Indian J Surg*. 2011;73(1):28-31. [CrossRef]
5. Jackie C, Andrew W, Robert H, et al. Thyroglossal duct cysts: Anatomy, embryology and treatment. *Surg Radiol Anat*. 2013;35:875-881. [CrossRef]
6. de Tristan J, Zenk J, Künzel J, et al. Thyroglossal duct cysts: 20 years' experience (1992–2011). *Eur Arch Otorhinolaryngol*. 2015;272(9):2513-2519. [CrossRef]
7. Alpay HC, Kaygusuz I, Karlidag T, Keles E, Yalcin S, Dabak H. Tiroglossal duktus kist ve fistülleri: 32 vakalık bir inceleme. *Firat Tip Dergisi*. 2007;12:287-289.
8. Mondin V, Ferlito A, Muzzi E, et al. Thyroglossal duct cyst: Personal experience and literature review. *Auris Nasus Larynx*. 2008;35(1):11-25. [CrossRef]
9. Açıkalın RM, Hacı C, Bayram AA, Gezginadam Z. Tiroglossal duktus kist ve fistüllerindeki klinik sonuçlarımız. *Med Bull Haseki*. 2016;54:94-96. [CrossRef]
10. Alatsakis M, Drogouti M, Tsompanidou C, et al. Invasive thyroglossal duct cyst papillary carcinoma: A case report and review of the literature. *Am J Case Rep*. 2018;19:757-762. [CrossRef]
11. Gioacchini FM, Alicandri-Ciufelli M, Kaleci S, et al. Clinical presentation and treatment outcomes of thyroglossal duct cysts: A systematic review. *Int J Oral Maxillofac Surg*. 2015;44:119-126. [CrossRef]
12. Sistrunk WE. The surgical treatment of cysts of the thyroglossal tract. *Ann Surg*. 1920;71(2):121-122.2.
13. Oomen KP, Modi VK, Maddalozzo J. Thyroglossal duct cyst and ectopic thyroid: Surgical management. *Otolaryngol Clin North Am*. 2015;48:15-27. [CrossRef]

14. Righini CA, Hitter A, Reyt E, et al. Thyroglossal duct surgery: Sistrunk procedure. *Eur Ann Otorhinolaryngol Head Neck Dis*. 2016;133:133-136. [CrossRef]
15. Ren W, Zhi K, Zhao L, et al. Presentations and management of thyroglossal duct cyst in children versus adults: A review of 106 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2011;111(2):e1-e6. [CrossRef]
16. Simon LM, Magit AE. Impact of incision and drainage of infected thyroglossal duct cyst on recurrence after sistrunk procedure. *Arch Otolaryngol Head Neck Surg*. 2012;138(1):20-24. [CrossRef]
17. Pradeep PV, Jayashree B. Thyroglossal cysts in a pediatric population: Apparent differences from adult thyroglossal cysts. *Ann Saudi Med*. 2013;33(1):45-48. [CrossRef]
18. Abuabara A, Baratto Filho F, Fuzza RF. Thyroglossal duct cyst. *South Braz Dent J*. 2010;7(2):244-246.
19. Öztürk K, Yaman H, Akbay E, Keleş B, Arbağ H, Özer B. Tiroglossal kist cerrahi sonuçlarımız. *Genel Tıp Dergisi*. 2005;15:117-120.
20. Rohof D, Honings J, Theunisse HJ, et al. Recurrences after thyroglossal duct cyst surgery: Results in 207 consecutive cases and review of the literature. *Head Neck*. 2015;37(12):1699-1704. [CrossRef]
21. Tarcoveanu E, Niculescu D, Elena CA, et al. Thyroglossal duct cysts. *J Chirurgie Iasi*. 2009;5(1):45-51.
22. Islam O. Thyroglossal duct cyst imaging. 2013. [CrossRef]
23. Yaman H, Durmaz A, Arslan HH, Ozcan A, Karahatay S, Gerek M. Thyroglossal duct cysts: Evaluation and treatment of 49 cases. *B-ENT*. 2011;7(4):267-271.
24. Yalçın Ş. Boyun kitleleri. In Çelik O (ed.): *Kulak Burun Boğaz Hastalıkları ve Baş Boyun Cerrahisi*. İstanbul: Turgut Yayıncılık, 2002:860-889.
25. Gupta P, Maddalozzo J. Preoperative sonography in presumed thyroglossal duct cysts. *Arch Otolaryngol Head Neck Surg*. 2001;127:200-202. [CrossRef]
26. Bsoul SA, Flint DJ, Terezhalmy GT, et al. Thyroglossal duct cyst. *Quintessence Int*. 2003;34:156-157.
27. Adeniran JO, Durell J, Lakhoo K. Neck: Cysts, sinuses, and fistulas. In *Pediatric Surgery*. Cham: Springer, 2020:395-404.
28. Flageole H, Laberge JM, Ngyuyen LT, et al. Reoperation for cysts of the thyroglossal duct. *Can J Surg*. 1995;38:225-229.
29. Ein SH, Shandling B, Stephens CA, et al. The problem of recurrent tyhroglossal duct remnants. *J Pediatr Surg*. 1984;19:437-439. [CrossRef]
30. Dedivitis RA, Camargo DL, Peixoto GL, et al. Thyroglossal duct: A review of 55 cases. *J Am Coll Surg*. 2002;194:274-277. [CrossRef]