

Thyroglossal duct surgery. What is the acceptable recurrence rate?

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ABSTRACT

Objectives: To present experiences of different specialties in the treatment of thyroglossal duct cysts (TGDCs) and subsequent complications in multiple centers.

Methods: A retrospective cross-sectional study of all cases of TGDC for a period of 11 years from 2008-2019 by different departments from 3 different centers in Jeddah, Kingdom of Saudi Arabia (King Faisal Specialist Hospital & Research Centre, Bakhsh Hospital and International Medical Center).

Results: Forty-nine patients were included. The type of surgery performed plays a significant role in recurrence ($p < 0.001$). The Sistrunk procedure had a lower recurrence rate (0%) than simple excision (70%) and has showed a significantly long recurrence-free interval ($p < 0.001$). Higher recurrence rates are associated with higher postoperative complications ($p = 0.002$). Patients who underwent pre-operative fine needle aspiration did not have any recurrence during the follow-up period.

Conclusion: The Sistrunk procedure is the gold standard technique with the highest recurrence-free interval rate. Fine needle aspiration could be recommended as a less invasive procedure to exclude malignancy.

Keywords: thyroglossal duct cyst, recurrence, sistrunk, surgical

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The thyroglossal duct cyst (TGDC) is one of the most common congenital midline neck masses that are located at the level of or below the hyoid bone in approximately 80% of cases, and the remaining 20%

are located above the hyoid bone.¹ Epidemiologically, TGDC has a bimodal distribution in children and young adults.² The clinical presentation of TGDC includes swelling, pain or tenderness, dysphagia, and dysphonia.³ However, inflammation or infection in the form of an abscess or cellulitis is the most common complication seen in TGDC patients. Other complications include the presence of the tract posterior to the hyoid bone or ectopic thyroid follicles.⁴ However, adults have been reported to experience more symptoms than children.²

Surgical removal is the definitive treatment; however, recurrence is a known possible outcome.³ To reduce the recurrence risk based on embryological principles, Schlange had suggested removal of the middle third of the hyoid bone with the main cyst; then, Sistrunk improved this technique further by removing the mid-portion of the hyoid bone and tissue between the hyoid bone and foramen cecum.⁵ However, the recurrence rate is still 6.6% in the pediatric population, irrespective of the type of surgery performed whether it is Sistrunk's operation or modified Sistrunk's procedure.⁶ Over time, it was found that the central neck dissection or anterior wide local excision to remove the entire thyroglossal tract remnant reduced the risk of initial failure and was considered a favorable option for the management of recurrent cases, especially in cases with a history of infected cyst or incision to avoid the risk of further recurrences.⁷

Few studies have emphasized the factors that are associated with TGDC recurrence that could increase the economic burden of the hospital and negatively influence the mental health of the patients and their family. The purpose of this study was to determine the factors related to TGDC recurrence and present the experiences of different specialties in the treatment of TGDCs and its complications in multiple centers.

Methods. A retrospective review of paper and electronic records were performed for all cases of TGDC for a period of 11 years from 2008 to 2019 by different departments (otolaryngology - head and neck surgery, pediatric surgery, and general surgery) from 3 different centers in Jeddah, Kingdom of Saudi Arabia (King Faisal Specialist Hospital & Research Centre, Bakhsh Hospital, and International Medical Center).

We included all patients with the diagnosis of TGDC without restriction to age, nationality, or presenting symptoms. We excluded patients with missing documentation and also those who were lost to follow up of <12 months. Data extracted from patients' records included basic demographic characteristics, clinical presentation, medical history, postoperative variables,

possible outcome parameters, and complications. All patients were followed up for 24 months after the surgery.

The literature review was carried out using PubMed, Medline, Embase, and Cochrane Library. Key terms related to thyroglossal duct cyst, recurrence, and Sistrunk were used to search for related published studies.

An institutional review board has approved this retrospective study and attests to its scientific validity. Informed written consent was provided by all participants in this study.

Statistical analysis. All statistical analyses were performed by the Statistical Package for Social Sciences for windows, version 25.0 (IBM Corp, Armonk, NY, USA). Descriptive statistics were formed from the data collected and expressed as mean, standard deviation (SD), and frequencies. Statistical comparisons between categorical variables were performed using Fisher's exact test or Chi-square test as appropriate. Kaplan-Meier (KM) curves were used to estimate the recurrence-free rate after surgery for TGDC. Moreover, a log rank test was performed to compare the recurrence-free rate among different procedures and specialists. A $p < 0.05$ was considered significant.

Results. A total of 49 patients were included in the study; of these, 32.7% were male. The mean±SD age of all patients was 11.8±11.4, and most of them were Saudis (83.7%). Only 5 patients (10.2%) presented with compression symptoms, and 14 patients (28.6%) had other comorbidities. Noteworthy, only 7 patients (14.3%) had a previous surgery, which was in a different hospital than the ones included in our study and had no records, for the thyroglossal cyst, and most of them were male (71.4%). Meanwhile, a total of 2 patients with papillary thyroid carcinoma (PTC) were reported, a 21-year-old male and a 22-year-old female. There was no significant difference in the examined characteristics between men and women (Table 1).

Association of recurrence with preoperative and operative variables. All patients with recorded recurrence (n=7) have been diagnosed with concomitant TGDC, and the cysts were situated either in the infrahyoid (71.4%) or the suprahyoid (28.6%). Regarding investigation modality, all patients

Table 1 - Basic sociodemographic characteristics of the included patients.

Variables	Gender		P-value
	Female	Male	
<i>Age (years)</i>			
0-4	4 (25.0)	6 (18.2)	0.979
5-8	3 (18.8)	6 (18.2)	
9-12	3 (18.8)	6 (18.2)	
13-16	1 (6.3)	3 (9.1)	
>17	5 (31.3)	12 (36.4)	
<i>Nationality</i>			
Non-Saudi	1 (6.3)	7 (21.2)	0.184
Saudi	15 (93.8)	26 (78.8)	
<i>Compression symptoms</i>			
No	15 (93.8)	29 (87.9)	0.524
Yes	1 (6.3)	4 (12.1)	
<i>Comorbidities</i>			
No	14 (87.5)	21 (63.6)	
Yes	2 (12.5)	12 (36.4)	
<i>Previous surgery for the thyroglossal cyst</i>			
No	14 (87.5)	28 (84.8)	0.804
Yes	2 (12.5)	5 (15.2)	

Values are presented as numbers and percentages (%).

underwent ultrasound preoperatively. Moreover, no recurrence was detected in patients with a history of fine needle aspiration (FNA) or in those who had their surgery performed by a general surgeon. There was no significant effect for any of the pre-operative variables on the recurrence rates (Table 2).

For the operative factors, surgery duration (mean±SD) was slightly longer in the recurrence group (90±43.8 min) than that in the no-recurrence group (85±59.5 min). In addition, a total of 5 surgeons with an average minimum experience of >15 years performed the surgeries. Additionally, the patients who underwent a resection of the middle (central) part of hyoid bone showed no recurrence. The general surgery group had no recurrence rate in comparison to the pediatric group which was 13.3% while the otolaryngology - head and neck surgery group had the highest rate of 23.1%. Nevertheless, there was no statistical significance found when comparing the data to those of the no-recurrence group (Table 2).

Association of recurrence with postoperative variables. All recurrent cases underwent a simple excision. A significant increase in risk of recurrence ($p < 0.001$) was found between simple excision and other procedures. Moreover, the recorded postoperative complications were significantly ($p = 0.002$) higher in the recurrence group (71.4%) than in the non-recurrence group (11.9%). However, none of the other postoperative variables have showed any significant difference between the recurrence and no-recurrence groups (Table 3).

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Recurrence-free rates (procedure and specialty). For both combined Sistrunk's and total thyroidectomy and Sistrunk's alone, the KM curve showed a similar recurrence-free rate (100%) during the follow-up period. In contrast, the simple excision procedure showed a significantly higher rate of recurrence ($p<0.001$) according to the log-rank test.

In terms of the specialty performing the surgery, general surgery showed superiority, with no recurrent cases recorded in their sample. However, no statistical significance was found for this difference ($p=0.202$) according to the log-rank test.

Discussion. This retrospective multicenter study describes our experience in TGDC management. Our study included 49 patients who underwent surgery for recurrent TGDC with histologically confirmed TGDC diagnosis in 86% ($n=42$) of the patients or other possible differential diagnoses, such as dermoid cyst in 8% ($n=3$), multinodular goiter in 2% ($n=1$), PTC in

2% ($n=1$), or lymph node (LN) with metastasis in 2% ($n=1$) of cases.

Thyroglossal cyst recurrence is common and depends mainly on the type of surgery and the incomplete removal of TGDC. In a retrospective study with a large sample size of 207 patients, the overall recurrence rate was 9.7%, and a significant difference was found among surgical types; the Sistrunk operation had a recurrence rate of 5.3%, which was lower compared to that of plain excision at 55.6%.⁸ In another retrospective review on a large number of patients ($n=352$), the overall recurrence rate was 4.5%.⁹ By using a 3-dimensional reconstruction, it was shown that TGDC penetrated the hyoid bone as a result of the forward growth of the hyoid bone. This could possibly result in recurrence.¹⁰ Dissection of all tracts is recommended to decrease the risk of recurrence; however, dissection of the foramen cecum was not found to be as important as the partial dissection of the hyoid bone.⁹

Furthermore, central neck dissection is considered

Table 2 - Preoperative and operative variables stratified by incidence of recurrence.

Variables	Recurrence			P-value*
	No	Yes	Total	
Consistency				
Cystic	31 (73.8)	7 (100)	38 (77.6)	0.124
Firm	11 (26.2)	0	11 (22.5)	
Location				
Lingual	1 (2.4)	0	1 (2.0)	0.893
Suprahyoid	10 (23.8)	2 (28.6)	12 (24.5)	
Infrahyoid	31 (73.81)	5 (71.4)	36 (73.5)	
Prophylactic antibiotics				
No	8 (19.1)	1 (14.3)	9 (18.4)	0.619
Yes	34 (81.0)	6 (85.7)	40 (81.6)	
Pre-operative diagnosis				
TGDC	38 (90.5)	7 (100)	45 (91.8)	0.528
TGDC with PTC	4 (9.5)	0	4 (8.2)	
Fine needle aspiration				
No	37 (88.1)	7 (100)	44 (89.8)	0.446
Yes	5 (11.9)	0	5 (10.2)	
Prophylactic antibiotics				
No	8 (19.1)	1 (14.3)	9 (18.4)	0.763
Yes	34 (81.0)	6 (85.7)	40 (81.6)	
Specialty				
Otolaryngology - head & neck surgery	10 (23.8)	3 (42.9)	13 (26.5)	0.592
General surgery	6 (14.3)	0	6 (12.2)	
Pediatric surgery	26 (62.0)	4 (57.1)	30 (61.2)	
Surgical duration (mean±SD)	85±59.53	90±43.79	86±57.18	0.835 [†]
Resection of the middle (central) part of hyoid bone				
No	3 (7.1)	0	3 (6.1)	0.623
yes	39 (92.9)	7 (100)	46 (93.9)	
Re-approximation of the hyoid bone				
No	13 (31.0)	3 (42.9)	16 (32.7)	0.412
Yes	29 (69.1)	4 (57.1)	33 (67.4)	

Values are presented as numbers and percentages (%). TGDC: thyroglossal duct cyst, PTC: papillary thyroid carcinoma, SD: standard deviation, *Fisher's exact test, [†]independent samples t-test

Table 3 - Postoperative variables stratified by the incidence of recurrence.

Variables	Recurrence			P-value*
	No	Yes	Total	
<i>Treatment</i>				
Sistrunk procedure	35 (83.3)	0	35 (71.4)	<0.001 [‡]
Simple excision	3 (7.1)	7 (100)	10 (20.4)	
Sistrunk procedure + total thyroidectomy	4 (9.5)	0	4 (8.2)	
<i>Drain placement</i>				
No	35 (83.3)	5 (71.43)	40 (81.6)	0.381
Yes	7 (16.7)	2 (28.57)	9 (18.4)	
<i>Histopathology</i>				
Confirmed diagnosis	35 (83.3)	7 (100)	42 (85.7)	1
Dermoid cyst	3 (7.1)	0	3 (7.1)	
Inflamed dermoid cyst	1 (2.4)	0	1 (2.0)	
MNG, no malignancy, TGDC	1 (2.4)	0	1 (2.0)	
PTC, absence of TGDC	1 (2.4)	0	1 (2.0)	
Level 1 L.N with metastatic PTC or TGDC	1 (2.4)	0	1 (2.0)	
<i>Postoperative complication</i>				
Fistula managed with antibiotics	2 (4.76)	2 (28.57)	4 (8.2)	0.002 [†]
Hematoma requiring surgical drainage	0 (0)	1 (14.29)	1 (2.0)	
Keloid scar	0 (0)	1 (14.29)	1 (2.0)	
None	37 (88.1)	2 (28.57)	39 (79.6)	
Postoperative infection managed with antibiotics	3 (7.14)	1 (14.29)	4 (8.2)	
<i>Postoperative antibiotic</i>				
None	2 (4.76)	0	2 (4.2)	0.827
Augmentin	24 (57.14)	4 (57.2)	28 (57.1)	
Cefalexin	7 (16.67)	2 (28.6)	9 (18.4)	
Cefazolin	3 (7.14)	0	3 (6.1)	
Cefuroxime	4 (9.52)	0	4 (8.2)	
Clindamycin	2 (4.76)	1 (14.3)	3 (6.1)	

Values are presented as number and percentages (%). MNG: multinodular goiter, TGDC: thyroglossal duct cyst, PTC: papillary thyroid carcinoma, L.N: lymph node, *Fisher's exact test, [†] $p < 0.05$, [‡] $p < 0.001$

an alternative method in decreasing the recurrence rate. Isaacson et al,⁷ reported that central neck dissection could reduce the risk of initial failure in difficult cases, and even though it is a safe procedure, it should still be performed with caution to avoid the risk of injury to the carotid artery, vagus nerve, or larynx.

However, there are other risk factors that could increase the risk of recurrence. The rupture of the cyst could increase the risk of recurrence; however, in another study, the rupture of the cyst in 53 out of 159 cases was noted to have no effect on recurrences or postoperative complications, despite the extension of the cyst posterior to the hyoid.^{3,11} Furthermore, postoperative infection may increase the risk of recurrence.⁸ Other possible factors could be attributed to the recurrence, such as expertise of the surgeon and years of training, the persistence of infra or suprahyoid tract remnants, and a misdiagnosis of a TGDC.¹²

Furthermore, our result revealed that only 2% were carcinomas or lymph nodes with metastasis. This finding is consistent with that of literature reporting that carcinoma could be present in approximately 1% of cases, but these cases had excellent prognosis if provided with adequate treatment.¹³ Moreover, in a retrospective

analysis, PTC in the epithelium of the cyst was reported in 1.4% of cases.⁹ However, FNA is still favorable to confirm the diagnosis and distinguish malignant features; it was carried out in 10.2% of our cases.¹⁴ Contrary to malignant transformation, the incidence of ectopic thyroid in TGDC is unknown. Thus, preoperative imaging, such as sonography, computed tomography, and FNA biopsy are recommended as supplementary techniques to confirm the diagnosis.¹⁵ In our study, we did not identify any patient with ectopic thyroid tissue in the postoperative histopathological examination which other studies demonstrated as commensurate finding.⁹⁻¹¹

Complications of the Sistrunk procedure are known to be minor ones and wound-related.¹⁵ In our study, we report no complications in 30/40 of patients who underwent Sistrunk operation, and in 39 out of the 49 patients in the total cohort. Moreover, other complications were related to infection and no statistically significant difference was found among the different types of surgery. In addition, the study has shown that surgeries conducted by general surgeons had a better prognosis as no recurrences were detected in cases that underwent general surgeries. These results

are consistent with another retrospective study of 102 patients who were followed up for 14 years.¹⁵ The study showed that there was no recurrence detected in the general surgery group as compared to the 3% recurrence rate found in the pediatric surgery group during the follow-up period.¹⁵ It is noteworthy that the pediatric surgery group (n=67) was larger than the general surgery group (n=35). Similarly, in our study, we only had 6 patients in the general surgery group, as compared to the 13 patients in the otolaryngology - head & neck surgery group and 30 patients in the pediatric surgery group.

Study limitations. A small sample size with a short follow-up duration can make it hard to draw an efficient conclusion. Secondly, we included all age groups with no comparison regarding specific groups. Lastly, patients coming from 3 different centers and 3 different departments may give rise to variabilities in surgical techniques. Therefore, further multicenter studies with a larger sample size with more evaluation of the Sistrunk surgical procedure among all surgical specialties with focus on recurrent TGDC cases are strongly suggested.

In conclusion, there is a significant difference in the recurrence rates and postoperative complications among different types of surgeries. No significant superiority has been detected when comparing the different specialties in terms of recurrence rates. The Sistrunk procedure is the gold standard technique, showing a higher recurrence-free rate as compared to a simple excision. Fine needle aspiration is highly recommended as a non-invasive modality to exclude malignancy and improve outcomes.

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