Thyroglossal Cyst and Fistula: Surgical pitfalls and causes of recurrence

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Background: The thyroglossal duct cyst (TDC) results from a failure of complete obliteration of thyroglossal duct. It represents the most common type of developmental cyst seen in the neck region.
Objective: Evaluate our experience in the management of primary and recurrent thyroglossal cyst and fistula and to determine the role of pre- and postoperative infection as an important factor associated with thyroglossal duct recurrence after surgery.
Methods: During the period from January 2013- April 2014, in General and Pediatric Surgery Department, 50 patients with thyroglossal duct cyst (35 patients) and fistulae (15 patients) were diagnosed and treated. All records were reviewed for age and sex, diagnostic methods, surgical management and postoperative infection and recurrences.
Results: The recurrence rate was high in cases with infection occurred preoperative or postoperative.
Conclusion: Infection very important leading factor for recurrence of thyroglossal cyst.

Keywords:
Sistrunk operation, thyroglossal duct cyst, recurrent thyroglossal cyst and fistula

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Introduction

A thyroglossal cyst is the most common congenital anomaly in the neck [1]. The disease is frequently encountered in children and adolescents but may appear in adults as well [2]. It arises from a persistent patent unobliterated portion of the thyroglossal duct that descends from the foramen caecum of the tongue to the neck. It usually presents as a soft cystic midline neck swelling but may present with complications, the most significant being infection. Diagnosis is made mainly by clinical examination and ultrasound examination.

The treatment of thyroglossal cyst is carried out with the Sistrunk operation, which entails removal of the cyst, the track to the foramen caecum and the central portion of the hyoid bone [3]. The recurrence rate of the condition after Sistrunk operation is 5% and rises to 20% if the hyoid bone is not removed [4]. Management of infected thyroglossal duct cysts traditionally included only antibiotics if cellulitis is present, or treated with incision and drainage when an abscess forms [5].

Patients and methods

This was a prospective study of 50 patients with symptoms and signs of thyroglossal cysts and fistulas (Fig. 1), referred to the Department of General and Pediatric Surgery, Zagazig University, during the period between January 2013 and April 2014. All patients underwent Sistrunk operation. Thyroglossal duct cyst was found in 30 cases (60%) and thyroglossal fistula in 20 cases (40%).

Informed consent was obtained from the parents before enrollment in the study. An informed written consent in accordance with the hospital Ethical Committee was also obtained.

The patients in this study were seen, treated, and followed up by a team of general and pediatric surgeons.

Detailed history and medical records of all patients with a diagnosis of thyroglossal cyst disease treated at our department from January 2013 to April 2014 were reviewed.

Preoperative measures included a thorough medical checkup and fitness for general anesthesia, and all patients were investigated by means of routine preoperative investigation; neck ultrasound was the mainstay of imaging for all cases.

Inclusion criteria

The study included all cases with thyroglossal cyst and fistulae.

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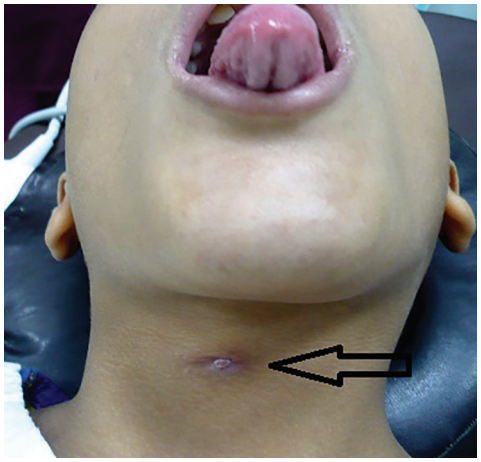
Figure 1



Thyroglossal cyst with fistula.

Figure 2

Transverse elliptical neck incision.

Exclusion criteria

Any case diagnosed intraoperatively with a diagnosis other than cyst or fistula was excluded, in addition to cases unfit for anesthesia. We excluded one case of midline cervical lymph node and one case of dermoid cyst.

Technique

All patients underwent general endotracheal intubation. The patients was made to lie supine with neck extension, and the neck was draped and prepped. Infiltration of the skin was carried out with lignocaine and adrenaline (1 : 200 000) and then the track of the fistula was delineated with methylene blue, followed by transverse elliptical skin incision in case of fistula (Fig. 2) or curved transverse incision in case of cyst. The subplatysmal space was reached and flaps were elevated (Fig. 3). The track was dissected with the removal of the central portion of the hyoid bone and V-shaped excision of the tongue was performed (Figs. 4 and 5). Proper hemostasis with a closure of the wound in layers was performed and the drain was inserted. The sutures were removed 1 week later and the patients were followed up from 6 months up to 1 year.

Parameters and data collection postoperatively

Standardized data collection was performed by the attending resident and our surgeon team, and each patient was evaluated by the main surgeon twice per day according to the hospital stay, and then the patients were followed up at the hospital outpatient clinic monthly for 6 to 12 months.

Follow-up data included patient age, sex, and clinical presentation, the presence of preoperative infection and postoperative infection, and recurrence.

Statistical analysis

Continuous variables were checked for normality using the Shapiro–Wilk test. Continuous variables were expressed as mean ± SD and median (range), and the categorical variables were expressed as number (percentage). Percentages of categorical variables were compared using the χ^2-test. All tests were two sided, with *P*-value less than 0.05 being considered statistically significant (S), *P*-value less than 0.01 considered highly statistically significant (HS), and *P*-value greater than 0.05 considered nonstatistically significant (NS). All data were analyzed using SPSS 22.0 for Windows (SPSS Inc., Chicago, Illinois, USA).

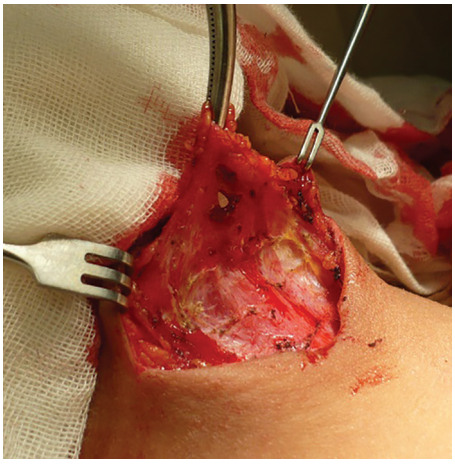
Results

One of the thyroglossal duct cysts was discovered histologically to be dermoid cyst and two cases were of prelaryngeal lymph nodes; the three cases were excluded from the study. A total of 35 patients had thyroglossal duct cyst preoperatively. The study group comprised 35 boys and 15 girls, with a sex ratio of 2.3 : 1. In the age group 3–7 years there were 25 patients (50%), in the age group 7–10 years there were 20 patients (40%), and the rest of patients were above 10 years of age, with a mean age of 8.1 years (Table 1).

The most common presenting complaints were a mass alone without infection (25 of 35 cases), mass with abscess or cellulitis (10 of 35 cases), and thyroglossal fistula without infection (15 of 50 cases). Most thyroglossal duct cysts were found in the classic midline infrahyoid position. However, four patients were noted to have their thyroglossal duct cysts in atypical locations: two were suprahyoid, one was infrahyoid and to the left, and one was infrahyoid and to the right (Table 2).

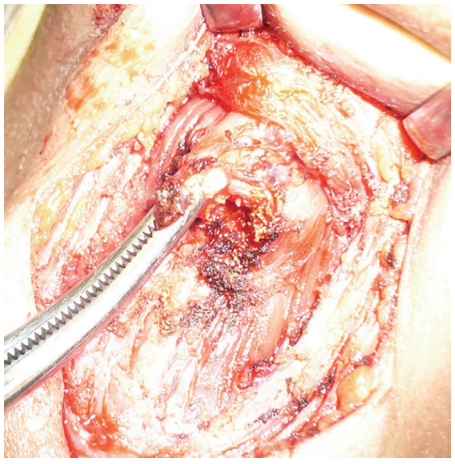
Data were analyzed in two ways. First, of the 35 patients who presented with thyroglossal duct cyst and underwent surgical intervention, there was no evidence of preoperative infection in 25 patients. In this group without preoperative infection, two patients had a recurrence of their disease, whereas 23 recovered uneventfully.

Figure 3



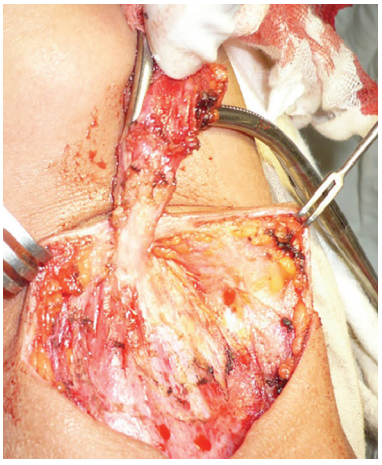
Subplatysmal dissection.

Figure 4



Excision of the central portion of the hyoid.

Figure 5



Removal of the central portion of the hyoid bone.

Of the 10 thyroglossal duct cyst patients who had a preoperative infection, three presented initially with an abscess and seven presented with cellulitis. In the three patients with preoperative abscess formation, drainage of the abscess was accomplished in patients (one incision and drainage and two spontaneously drained). In this group of preoperative abscess patients, two patients had recurrent disease. Of the seven patients who presented initially with cellulitis and underwent preoperative antibiotic therapy, two patients had recurrent disease.

There was a statistical difference with regard to recurrence between those patients who presented with infection (four of 10) and those who did not present with infection (two of 25) (Table 3).

The second way in which these data were analyzed was by comparing postoperative infection with the development of a recurrent disease. Postoperative infection developed in 10 patients of the 50 patients. In three patients, there was no evidence of recurrent disease, and only seven of these 50 (14%) patients had a postoperative recurrence, four of whom had a preoperative infection. Each of these seven patients with recurrent disease underwent reoperation with

Table 1 Demographic data

Demographic data	*n* (%)
Age (years)	
Mean ± SD	8.1 ± 2.2
Median (range)	8 (3–13)
3–7 years	25 (50)
>7–10 years	20 (40)
>10 years	5 (10)
Sex	
Male	35 (70)
Female	15 (30)

Table 2 Complaint and site of cyst or fistula

Complaint and site of cyst or fistula	*n* (%)
Complaint	
Thyroglossal duct cyst	35 (70)
Without infection	25 (50)
With infection	10 (20)
Thyroglossal fistula	15 (30)
Site of cyst or fistula	
Infrahyoid midline	46 (92)
Infrahyoid to right	1 (2)
Infrahyoid to left	1 (2)
Suprahyoid	2 (4)

wide excision of the area and re-excision of the hyoid bone (Table 4).

Discussion

Thyroglossal cyst disease is the most common developmental neck lesion in the pediatric group. There is a slight male predominance. These lesions usually present as a midline mass, but can present initially as a draining sinus or abscess [6,7]. Most thyroglossal duct cysts present during the first 5 years of life, although the lesion has been repeatedly described during adulthood [8]. Associated ectopic thyroid tissue was found in one case only in our specimen, but thyroid gland was in normal place. An author studied the recurrence rate after Sistrunk operation and reported a case with an intrathyroidal thyroglossal cyst [9].

In this study, preoperative infection was detected in 10 cases: seven cases were of cellulitis and three cases were of abscess, which was treated with incision and drainage. Another study [10] stated that only one patient in their study (5%) had a history of infection and the culture showed Homophiles influenza.

In this study, there were seven recurrent cases and most of them were due to postoperative infection. Infection may cause rupture of the cyst and implantation of the epithelium of the cyst into the surrounding tissue and is responsible for recurrence. Recurrence rate of thyroglossal cyst after complete excision using the Sistrunk procedure is reported to be 2.6–5%, whereas simple excision of the cyst can result in recurrence rates as high as 38–70%. Other authors [11,12] have reported a recurrence in two cases in a series of 62 patients. Ein *et al*. [13] reported a recurrence rate of 10% in a series of 270 patients, with most recurrences occurring when the middle third of the hyoid was left intact.

Authors [14,15] have reported a recurrence rate of 3.4% in their series of 29 patients who underwent the Sistrunk procedure. Recurrence rates ranging from 1 to 30% have been reported in a few other series [16,17]. The most common cause of recurrence is rupture of the cyst intraoperatively or leaving a part of the wall behind.

Histological analysis of the specimens showed that there were three cases misdiagnosed preoperatively as thyroglossal cyst, but histopathological examination postoperatively showed two cases of lymph nodes and one case of dermoid cyst. Athow *et al*. [18] stated that dermoid cysts, second in incidence to thyroglossal duct cyst, are reported to sometimes occur simultaneously with thyroglossal duct cyst.

Table 3 Preoperative infection and recurrence

Preoperative infection and recurrence	*n* (%)
Preoperative infection	10 (20)
Abscess	3 (6)
Cellulitis	7 (14)
Recurrence	6 (12)
Presence of preoperative infection	4 (8)
Absence of preoperative infection	2 (4)
Test[a]	5.149
P-value	0.023

[a]The value is calculated using the χ^2-test; $P < 0.05$, significant.

Table 4 Postoperative infection and recurrence

Postoperative infection and recurrence	*n* (%)
Postoperative infection	10 (20)
Recurrence	7 (14)
Presence of preoperative infection	4 (8)
Absence of preoperative infection	3 (6)

Conclusion

To prevent recurrence of thyroglossal cyst and fistula, complete excision of the track and central portion of the hyoid bone should be performed.

Infection is a very important factor preoperatively and postoperatively that increases the risk of recurrence of thyroglossal fistula.

Postoperative infection is associated with a higher rate of recurrence compared with preoperative infection.

Recommendation

Sistrunk operation is performed as an ideal surgery for the treatment of thyroglossal cyst and fistula.

Every effort is taken to prevent postoperative infection as it is the important factor causing recurrence of the disease.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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