Evaluation of Surgical Outcomes and Causes of Recurrence in Thyroglossal Duct Cyst in Referral Children Hospital of Iran from 2011 to 2018

Leily Mohajerzadeh[1], Sadaf Yazdani[1]*, Mehdi Sarafi[1], Ahmad Eghbali[1], Amir Mohammad Zakeri[1]

[1] Pediatric Surgery Research Center, Research Institute for Children's Health, Shahid Beheshti University of Medical Sciences, Tehran, Iran

*Address for Corresponder: Dr. Sadaf Yazdani, Pediatric Surgery Research Center, Research Institute for Children's Health, Shahid Beheshti University of Medical Sciences, Tehran, Iran. (email: sfyazdani@gmail.com)

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Abstract

Introduction: Thyroglossal duct cyst is amongst the most common congenital anomalies in the neck region which is only properly treated with surgery. However, the long-term result remains uncertain. The present study aimed to assess surgical outcomes and predictors of recurrence in cases of thyroglossal duct cyst that have been referred to a tertiary level children hospital in Iran.

Materials and Methods: In this cross-sectional study, 29 children with thyroglossal duct cyst that had undergone Sistrunk procedure in Mofid hospital between 2011 and 2018 were evaluated. The study information was collected by reviewing the hospital charts.

Results: Patient's age ranged between 20 and 40 months. The disease in boys was twice as prevalent as girls and hence male to female ratio was almost equal to 2 to 1. Palpable cervical mass was the most prevalent which we found in 62.1%, followed by painful neck mass (13.8%), discharging cutaneous fistula (13.8%), palpable mass with dysphagia (6.9%) and abscess (3.4%). Preoperative antibiotics were prescribed for infection in 34.5%. Regarding the position of the thyroglossal duct cyst, suprahyoid position was reported in 72.4%. In terms of type of surgery, 23 cases (79.3%) underwent sistrunk procedure and 6 cases (20.7%) underwent simple incision. The mean follow-up time for patients was 3.32 ± 1.42 years. The relapse rate after surgery was 6.9% and the rate of postoperative complications was 10.3%. The percentage of relapse-free survival was 91.3% during the follow up period. The presence of discharging fistula as the predominant manifestation (p = 0.009), preoperative infection (p = 0.043), and thyrohyoid position (p = 0.016) were related to the disease recurrence.

Keywords

- Recurrence
- Thyroglossal cyst
- Congenital neck mass

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Conclusion: Surgical complications rate including relapse or infections was 10.3% and the incidence of recurrence after surgery during the follow-up period with a mean of three years was 6.9%; that is more prevalent in patients with the presence of discharging fistula, preoperative infection, or thyrohyoid position. Preoperative infections, rather than postoperative infections, are associated with more recurrence. Thus infection is very important factor in the recurrence of thyroglossal duct cyst.

Introduction

Thyroglossal duct cyst is one of the most common congenital anomalies in the neck region, accounting for about 7% of the population.[1,2] This lesion represents about 2 to 4 percent of the total cervical mass and in total 70 percent of congenital neck abnormalities. In terms of diagnostic approach, approximately three-quarters of the thyroglossal duct cyst are cystic, while 25% are in the form of drainage sinuses and fistulas.[3] The sinuses are the result of an infection (in the cyst) and sometimes accompanied by cystic tears that will drain out. Thyroglossal duct cysts are mostly asymptomatic and sometimes seen as a local swelling on the skin of the hyoid bone.[4] In terms of therapy, surgical removing the cysts is the gold standard therapeutic approach. This will be done by removing the center of the hyoid bone, which is called the Sistrunk surgery.[5] Approximately 10% of cysts may remain and need a second repairing surgery for removing remnants.[6,7] In patients infected or accompanied by drainage of the contents of the cyst to the outside, recurrence is likely to occur.[8,9] In some recent studies, the most commonly reported post-surgical complication was local infection (in 4% of cases), and overall recurrence has been reported to be about 11%.10 The best treatment with the least relapse rate is the same as the Sistrunk procedure,11-13 however in recent years the use of minimally invasive methods has been very much considered. What we did in this study was to assess surgical outcomes and causes of recurrence in thyroglossal duct cyst referred to Mofid children hospital between 2011 and 2018.

Materials and Methods

In this cross-sectional study, 29 children with thyroglossal duct cyst that had undergone Sistrunk procedure in Mofid hospital between 2011 and 2018 were evaluated. For patients, a checklist including patient information as demographic data, clinical manifestations, results of Sistrunk surgery and its related complications was collected and recorded by reviewing the patients' hospital recorded files. Finally, the information was entered into a statistical file and analyzed using the SPSS software.

Results

In this study, a total of 29 children with thyroglossal duct cyst that had undergone surgery in Mofid Children's Hospital between 2011 and 2018 were evaluated. The baseline characteristics are summarized in **Table 1**.

Table 1: Baseline characteristics of study population

Mean age, month	**41.93 ± 23.70**
Sex	
Male	62.1%
Female	37.9%
Mean admission time, day	1.90 ± 1.56
Preoperative antibiotic use	34.5%
Imaging studies	
Ultrasonography	51.7%
Ultrasonography with isotope thyroid scan	48.3%
Position of duct	
Suprahyoid	72.4%
Thyrohyoid	17.2%
Suprasternal	6.9%
Infralingual	3.4%
Type of surgery	
Classic sistrunk	79.3%
Simple incision	20.7%

The mean age of children was 41.93 ± 23.31 months from 14 to 92 months. In terms of the manifestation of the disease, palpable cervical mass was the most prevalent seen in 18 cases (62.1%), followed by painful neck mass in 4 cases (13.8%), discharging cutaneous fistula in 4 cases (13.8%), palpable mass with dysphagia in 2 cases (6.9%) and abscess in 1 case (3.4%). In terms of the pre-surgical treatment approach, antibiotics were prescribed for infection in 10 cases (34.5%). In terms of diagnostic imaging, 15 cases (51.7%) were evaluated by ultrasonography and 14 (48.3%) were evaluated by ultrasonography with scan radioisotope thyroid. In terms of type of surgery, 23 cases (79.3%) underwent sistrunk procedure and 6 cases (20.7%) underwent simple incision. In the postoperative diagnosis, the final diagnosis was thyroglossal duct cyst in 25 (86.2%) cases, and dermoid cyst in 4 cases (13.8%). Also, in pathological assessment, the final diagnosis was thyroglossal duct cyst in 17 cases (58.6%), dermoid cyst in 11 cases (37.9%) and atypical mycobacterial infection in 1 case (3.4%).

The mean follow-up time for patients was 3.32 ± 1.42 years (range 1.5 to 8 years). In terms of surgical outcomes, recurrence occurred in 2 (6.9%) cases, and in 1 case (3.4%), post-surgical infection was observed that improved with antibiotic therapy. Accordingly, the relapse rate after surgery was 6.9% and the rate of postoperative complications

was 10.3%. Based on the survival rate calculation using the Kaplan-Meier survival curve **Figure 1**, the percentage of relapse-free survival was 91.3% during the follow up period, which was fully maintained by the end of the eighth year of the follow-up.

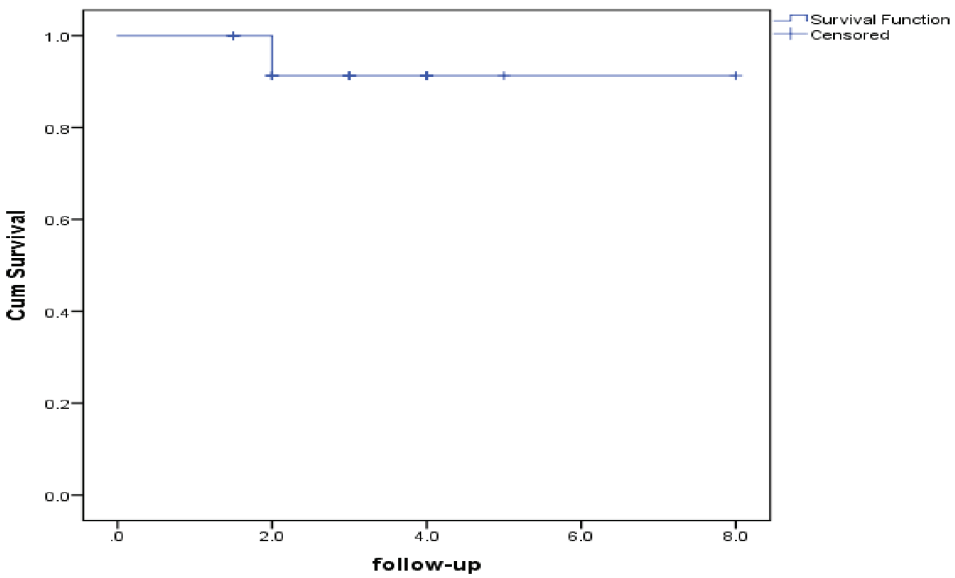

Figure 1: A relapse-free survival curve in patients with thyroglossal duct cyst

In the evaluation based on univariate analysis and among the underlying indices, the presence of discharging fistula as the predominant manifestation (p = 0.009), preoperative infection (p = 0.043), and thyrohyoid position (p = 0.016) was related to the disease recurrence. Accordingly, the frequency of relapse in patients with and without fistula secretion was 50% and 0%, in patients with and without preoperative infection was 20% and 0% respectively, and in patients with and without thyrohyoid position was 40% and 0% respectively. In this regard, the patient's gender (p value = 0.512), age (p value = 50.04), hospital admission time (p value = 0.411), or surgical type (value p = 0.445) was not related to the recurrence of the disease. Unfortunately, because of the limited sample size, it was not possible to use the Cox proportion hazard model to determine predictors of recurrence.

Discussion

One of the interesting results found in this study was that despite the confirmation of the thyroglossal duct cyst in the imaging methods, the cases with the ultimate diagnosis of the disease were 86.2% for surgery and 58.6% for the pathology. Initially, with regard to the epidemiological aspects of the disease, a review of the findings of other studies led to similar and sometimes contradictory results. In a recent systemic review study, 94% of patients were treated by sistrunk procedure, which was 80% in our study. In terms of clinical manifestations,

palpable neck mass was reported in 75%, abscess or cervical infection in 34% and dysphagia in 9%, which in our study was 62.1%, 3.4% and 6.9%, respectively; which was fundamentally different from our findings. In the corresponding meta-analysis, the complications of treatment were reported in 8% of cases, which in our study was reported to be 10.3%, which was somewhat close to our findings.14 In a study by Zaman et al in 2017, in terms of sex distribution, 71.4% were boys, which was similar to our study. In their study, common symptoms were the palpable cervical mass that was consistent with our study as well.15 In Thompson et al study in 2016; the most common clinical manifestation was mobile cervical mass in the midline of the neck was mainly in the infrahyoid position. In their study, the frequency of relapse was 3%, which is far less than the recurrence rate of our study.16 In a study by Ubayasiri et al in 2013, postoperative complications were reported in 11% of cases and the frequency of recurrence was 6%,17 which was very similar to our findings, considering that their study mostly included adults. Comparing the results of our study with others, we conclude that first; there is no difference in clinical manifestations and outcome of surgery in children and adolescents. Second, most patients have long term recoveries, although the relapse rate varies widely from 6% to 30%, which is, of course, influenced by the patient's age, follow-up time, and surgical technique. Of course, based on the final analysis in our study, three predictors of recurrence included the presence of discharging fistula, preoperative infection, and thyrohyoid position. This finding emphasizes that, firstly, antibiotic prophylaxis before surgery will significantly reduce the incidence of post-surgical recurrence. Secondly, the experience of surgery with the aim of repairing fistula, and especially in thyrohyoid position may be associated with higher successful operation as well as minimizing the recurrence.

Another important point in this study was the low sensitivity and specificity of imaging techniques, especially ultrasonography, to detect thyroglossal duct cyst. Considering the surgical findings, the positive predictive value of imaging for diagnosis of thyroglossal duct cyst was 86.2%, and compared to pathologic findings, it was 58.6%, which would be far lower. Therefore, in order to confirm the diagnosis, the diagnosis should be based on the findings of the pathology.

The study had some limitations. First, the limited sample size of the study did not allow us to determine the predictors of relapse by eliminating the confounding factors of our study. Second, due to the limited study volume and, as a result, the limited number of cases of relapse, it was not possible to determine the absolute relapse-free survival rate.

Conclusion

Surgical complications rate including relapse or infections was 10.3% and the incidence of recurrence after surgery during the follow-up period with a mean of three years was 6.9%; that is more prevalent in patients with the presence of discharging fistula, preoperative infection, or thyrohyoid position. Preoperative infections, rather than postoperative infections, are associated with more recurrence. Thus infection is very important factor in the recurrence of thyroglossal duct cyst.

Ethical Consideration

This study was approved by 36 sessions of Medical Ethics Committee of Shahid Beheshti University of Medical Sciences with Ethical Code Number of "IR.SBMU.MSP.REC.1395.477" on 95.11.12.

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Conflict of interests

There is no conflict of interests

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