ORIGINAL ARTICLE

Experience of Sistrunk's Operation for Paediatric Patients of Thyroglossal Duct Cyst at Multicenter Tertiary Care Hospitals

ABID HUSSAIN¹, SOOFIA MUSTAFA², MUHAMMAD FAROOQ³, ABDUL REHMAN⁴, HASAN TARIQ⁵, MUHAMMAD AFZAL MIRZA⁶

¹Assistant Professor of Paediatric Surgery, Khawaja Muhammad Safdar Medical College Sialkot

²Assistant Professor Paediatric Surgery, Bahawal Victoria Hospital Bahawalpur

³Assistant Professor of ENT Department, Shahida Islam Medical College Lodhran

⁴Senior Registrar Pediatric Surgery, Sialkot Medical College

⁵Senior Registrar Oral and Maxillofacial Surgery, Khawaja Muhammad Safdar Medical College, Allama Iqbal Memorial Teaching Hospital Sialkot

⁶Professor of Paediatric Surgery, Khawaja Muhammad Safdar Medical College Sialkot

Corresponding author: Dr Abid Hussain. Email: drabid381@gmail.com. Mobile: 0300 6831292

ABSTRACT

Background: The Thyroglossal duct cyst (TGDC) is a congenital defect resulting from a persistent thyroglossal tract. It usually exhibits as a painless nodule in the neck midline. In physical evaluation, it moves with swallowing and on tongue protrusion. The physical examination and detailed history are adequate to make an accurate diagnosis preoperatively. Neck ultrasound and Fine needle aspiration cytology are additional valuable diagnostic tools. Though it is a congenital abnormality, it usually appears in the 1st and 2nd decades of life. Sistrunk operation is the treatment of choice for cysts of the thyroglossal duct.

Aim: The purpose of this analysis was to govern the age at onset, site of cyst in relation to the hyoid bone, time of thyroglossal duct cysts presentation and to observe postoperative complications of Sistrunk's operation. **Study Design:** A Retrospective study.

Place and Duration: In different Departments like Paediatric Surgery, Otolaryngology, Faciomaxillary from January 2016 to January 2021.

Methods: A total of 76 patients operated in 5 years were included. The study included patients treated with the Sistrunk procedure in whom the presence of the thyroglossal duct was confirmed after clinical examination and ultrasonography. Demographics, age at admission, wound infection, hematoma, abscess, airway injury, seroma, stitch sinus, wound dehiscence and recurrence were observed.

Results: There was a slight high prevalence among men. Most of the thyroglossal cysts were seen in the pediatric inhabitants and below the hyoid bone and can recur.

Conclusion: Thyroglossal duct cyst is a pediatric disease. The Sistrunk procedure is the treatment of choice for TDC. The risk of serious complications is minimal when the procedure is carried out with certain nuances which we have described. The complications are minor and concerned to the wound.

Keywords: Recurrence, Position, Thyroglossal duct cyst and Sistrunk's operation.

INTRODUCTION

Thyroglossal duct cyst (TDC) is a congenital anomaly caused by persistent of thyroglossal duct. It is a very common neck swelling among children and very common clinical differential diagnosis in pediatric OPD1-2. While the site of the thyroglossal cysts can vary from intraoral to hyoid or infrahyoid position, mostly appear as nodules which may be painless under the hyoid bone in the neck³⁻⁴. The hoarseness and dysphagia never occur⁵. There are also case reports of papillary carcinoma of thyroid emerging inside the cyst⁶. On physical inspection, the cyst of the Thyroglossal duct moves together with the tongue protrusion (due to the connection of thyroglossal duct remnants with root of tongue) and with swallowing⁷⁻⁸. It is one of the utmost distinctive clinical signs that distinguishes it from various swellings of the neck like thyroid gland, dermoid cyst, lymph node, sublingual gland, etc. In many cases, the physical examination and detailed history are adequate to make an accurate diagnosis preoperatively. Fine needle aspiration cytology and neck ultrasound are additional valuable diagnostic tools9. Though it is a congenital abnormality, it usually appears in the 1st and 2nd decades of life. A bimodal distribution of age in TDC was noticed, with a highest incidence among 6-13 and ≥19

years of age. Sistrunk surgery is the preferred standard of treatment for cysts of the thyroglossal duct¹⁰. This operation involves the excision of the central part/body of the hyoid bone along with the cyst and its track up to the foramen cecum of tongue in order to prevent recurrence. Simple cyst excision is related with an augmented rate of recurrence¹¹⁻¹². The purpose of this analysis was to govern the age at onset, site of origin in relation with the hyoid bone in patients with thyroglossal duct cysts and to observe postoperative complications like wound hematoma, abscess, airway injury, seroma, stitch sinus, wound dehiscence and recurrence. We expect that this analysis will raise our treatment standards for thyroglossal duct cysts in terms of the presenting age of patients, the site of the thyroglossal cyst in association with the hyoid bone and postoperative complications related to Sistrunk surgery.

METHODS

This is a retrospective analysis conducted at the Paediatric Surgery Departments of Govt. Sardar Begum Teaching Hospital, Sialkot and BVH, Bahawalpur, Faciomaxillary Unit at AIMTH, Sialkot and Department of Otolaryngology, Shahida Islam Medical and Dental College and Teaching

Hospital, Lodhran for five-year duration from January 2016 to January 2021. The thyroglossal cyst diagnosis was based on clinical examination and neck ultrasound. A total of 76 patients operated in 5 years were included. The study included subjects of both genders and pediatric age group with proven thyroglossal cysts who underwent Sistrunk surgery. All specimens found to be TGDC confirmed by histopathology were included finally in the analysis. From the clinical files of hospitalized patients, the surgical record and follow-up of the patients were analyzed. The volunteers were called by phone regarding their postoperative status and asked for a follow-up visit if possible. Patients who visited minimum 6 months later to the respective departments were enrolled in the analysis. The patients of thyroglossal fistula or other diagnosis at the time of surgery were excluded from the study. The approval was obtained from the Hospital Ethical Review Board. Data were analyzed and collected for site of origin, recurrence of the cysts, age on presentation and the outcome was presented.

RESULTS

Over five-years, 76 patients with thyroglossal cysts were removed by Sistrunk surgery. All patients experienced the similar standard technique of surgery without bias. Our study included 40 men and 36 women with a male to female ratio of 1.11: 1. The mean age was 12.34 years. The patients < 10 years of age were (34) and then 10 to 16 years old (42).

Table-I shows the demographic features of the patients

40(52.6%)
36(47.4%)
12.34 ± 3.1
16(21.1%)
18(23.7%)
42(55.3%)

There were 42 patients over the age of 10. Patients above the age of 10 accounted for approximately 50% (55.3%) of the overall number of patients in this analysis. According to one sample t test; the age ranges have statistical significance (two-tailed P value 0.0020).

Table-IV. Comparison between the cyst size and presentation to nospital subsequently occurrence of the cyst				
	Maximum transverse diameter of the (millimeters)	cyst		
Presentation to the hospital after occurrence of the cyst	1-19 mm	>20 mm		
0-1 yr	24	14		
>1 yr	16	22		

In this technique, no significant complications were detected after the surgical intervention.

Table-V: Possible Complications of the Sistrunk Procedure

Recurrence	8	10.5%
hematoma	5	6.6%
Inadvertent entry into the airway	0	0
Seroma	4	5.3%
Local wound infection	5	6.6%
Stitch abscess	1	1.3%
Wound dehiscence	0	0

The localization of the cyst of the thyroglossal tract has also been noted. The suprahyoid position was found in 16 patients (21.1%) while the infrahyoid position in 60 (78.9%). Though, there was no statistically substantial variance amid the two sites (p-value, two tailed = 0.39). There was variable time of presentation from the appearance of the cyst to the date of admission in the hospital. 50% (38) of patients reported within one year of onset of neck swelling, 16 in the 2nd year, 12 after 2-5 years, and the remaining 10 later to the ten years. The mean time from the appearance of the cyst to the hospital admission was 4.20 years. (Table -II)

Table-II shows the duration from incidence of cyst to the hospital presentation and relationship with hvoid bone

presentation and relationship with hybrid bone		
Occurrence of cyst	No of Patients	
0-1 years	38	
1-2 years	16	
2-5 years	12	
> 10 years	10	
Hyoid Position		
Suprahyoid	16(21.1%)	
Infrahyoid	60(78.9%)	

There were also differences in the size of the cysts. 40(52.6%) of the cysts were small and had a maximum size less than 20 mm on ultrasound, 24(31.6%) had a maximum size from 20 to 29 mm, and 12(15.8%) ranged from 30 to 39 mm on ultrasound. None of the cysts were larger than 40 mm (Table-III).

Table-III shows the number of patients rendering to the cyst size

Cyst Size	No of patients
1-19mm	40(52.6%)
20-29mm	24(31.6%)
30-39mm	12(15.8%)

There was no significant relation between the increased cyst size and late presentation. 8 of the 76 cases had a postoperative relapse and the relapse rate was 10.5%. Six were treated conservatively with dressing and wide bore aspiration by needle while two individuals needed modification surgery.

Minor complications like seroma noticed in 4(5.3%) patients, haematoma in 5 (6.6%), local wound infection in five (6.6%) patients and stitch abscess occurred in one (1.3%) patient. Recurrence noticed in eight (10.5%) patients.

DISCUSSION

Congenital midline swellings of the neck are rare lesions of various clinical picture and embryogenesis. Diagnosis can very difficult for pediatric surgeons otolaryngologists.

A detailed medical history and clinical examination should be taken to make an accurate diagnosis before surgery. A cyst of the thyroglossal duct is a congenital abnormality that fallouts from the continuity of the thyroglossal duct as it descends from the foramen cecum to the normal position of the thyroid gland in the neck. In the phase of descent, the duct is mainly stuck in the hyoid bone, as the development of hyoid bone occurs from the lateral to the medial and its position also changes 12-13. The backbone treatment of the thyroglossal cyst is surgery. The prototype of the operation in this situation is the Sistrunk operation which purposes to eliminate the cyst along with the hyoid bone body between its 2 great cornu and some muscles above the hyoid bone upto foramen caecum.

Although it is one of the birth defects, a cyst of the thyroglossal duct is generally observed at birth. The maximum communal demonstration of TDC in this study was in the 1st ten years of life, trailed by the 2nd decade. Though, In Arun kumar et al study of 16 patients found that cysts often occur in the 2nd era of life¹⁴⁻¹⁵. 12.34 years was the patients mean age at presentation in this study. Comparable results were presented by other investigators in the various studies¹⁶⁻¹⁷. The position of the cyst in association with the hyoid bone is also significant for planning of the surgery. Cysts over the hyoid have less chances to relapse. In Sistrunk surgery, part of the hyoid bone is excluded as the duct passes in front of, behind, or through the hyoid bone. In our study, the suprahyoid position was found in 16 patients (21.1%), and the infrahyoid in 60 (78.9%). In Thompson et al study, involving 685 patients, the most common location was the infrahyoid location 18-19. In the Sarkar et al study; most of the cysts (83.3%) were located in the infahyoid, trailed by the suprahyoid in ten percent of cases. These studies are in line with this analysis. Comparable explanations were made by Debnath et al study20-21. In this analysis, no statistically significant relationship between presentation and increased cyst size was noted. This fact was not mentioned in the literature. In our study, the size of the thyroglossal cyst ranged from 10 mm to 39 mm in the largest dimension, and 52.6% has less than 20 mm cyst. However, a study by Patigaroo et al. Found that the largest size of most cysts is between 16 and 30 mm²¹. In one study, the duration of symptoms prior to admission ranged from 1 week to 6 years (median: 6 months). Serious complications after the Sistrunk procedure include relapse in 10.5% of patients, abscess or hematoma requiring surgery in 6.6%, no airway injury in any patient, local wound infection in 6.6% of patients and 1% of patients had a suture abscess²².

Ducic et al. found that the only statistically significant difference between the successful and unsuccessful results was the presence of a preoperative infection²⁶. Von Bismarck and Hollwarth found that patients with symptoms of infection or incomplete hyoid resection had a higher risk of recurrence and recommended early surgical treatment. On the other hand, Burgues Prades et al. study showed that there was no significant association between relapse and the preoperative TGDC inflammatory episode ^{23,25}.

The patient's number in this study was 76 with minimum inclination towards men. This is in line with the

work of Swaid et al. In their analysis, the M:F ratio was 2.3: 1

Cyst recurrence occurred in 8 of our patients (10.5%) after Sistrunk surgery. The recurrence rates in the Lawrence study and the Tom study were 10.8% and 11.5%, respectively²⁴

CONCLUSION

The Sistrunk procedure is the surgery of choice for the treatment of TDC because it has been associated with a low relapse rate in the past. The risk of serious complications is minimal. The complications are minor and concern the wound. Thyroglossal cysts are small to medium-sized swellings in the neck that move with both swallowing and tongue protrusion. It generally presents in the first decade of life with the bimodal presentation of age. The common location is Infrahyoid, there is no correlation amongst first decade of life and late demonstration, and the Sistrunk surgery is definite treatment with an acceptable chance of relapse.

REFERENCES

- Pucher B, Jonczyk-Potoczna K, Kaluzna-Mlynarczyk A, Kurzawa P, Szydlowski J. The central neck dissection or the modified sistrunk procedure in the treatment of the thyroglossal duct cysts in children: our experience. BioMed research international. 2018 Jun 19;2018.
- Koirala K. Sistrunk's operation: ten years' experience in a teaching hospital. Nepal Journal Of Medical Sciences. 2020 Feb 15;5(1):20-7.
- Zaman SU, Ikram M, Awan MS, Hassan NH. A decade of experience of management of thyroglossal duct cyst in a tertiary care hospital: differentiation between children and adults. Indian Journal of Otolaryngology and Head & Neck Surgery. 2017 Mar 1;69(1):97-101.
- Coelho A, Sousa C, Marinho AS, Barbosa-Sequeira J, Ribeiro-Castro J, Carvalho F, Moreira-Pinto J. Five-years' experience with outpatient thyroglossal duct cyst surgery. International journal of pediatric otorhinolaryngology. 2017 May 1:96:65-7.
- Mehtani N, Frauenfelder C, Rudd J, Hartley B. 107 The extended sistrunk procedure for thyroglossal duct cysts: a review of 168 cases.
- Alahmadi AA, Bawazir OA, Rajab MK, Althobaiti IA, Bawazir AO, Abi Sheffah FR, Al-Tammas AH, Marglani OA, Heaphy JC, Alherabi AZ. Thyroglossal duct surgery. What is the acceptable recurrence rate?. Saudi Medical Journal. 2020 Aug;41(8):878.
- Anuwong A, Jitpratoom P, Sasanakietkul T. Bilateral areolar endoscopic Sistrunk operation: a novel technique for thyroglossal duct cyst surgery. Surgical endoscopy. 2017 Apr 1;31(4):1993-8.
- Bakkar S, Biricotti M, Stefanini G, Ambrosini CE, Materazzi G, Miccoli P. The extent of surgery in thyroglossal cyst carcinoma. Langenbeck's archives of surgery. 2017 Aug;402(5):799-804.
- Qu R, Wang C, Dong Z, Li J, Liu D. Another strategy for the treatment of thyroglossal duct cyst: totally endoscopic surgery by breast approach. Surgical laparoscopy, endoscopy & percutaneous techniques. 2018 Apr 1;28(2):118-22.
- Sistrunk JW, Shifrin A, Frager M, Bardales RH, Thomas J, Fishman N, Goldberg P, Guttler R, Grant E. Clinical performance of multiplatform mutation panel and microRNA risk classifier in indeterminate thyroid nodules. Journal of the American Society of Cytopathology. 2020 Jul 1;9(4):232-41.

- Gao K, Han J, Zhou X, Luan D, Xie F, Li Y, Yue Z. A new transoral modality for the treatment of lingual thyroglossal duct cyst with suspension laryngoscopy by plasma coblation. The Annals of The Royal College of Surgeons of England. 2021 Jun;103(6):438-43.
- Hossain MS, Bhuyan MA, Kafi AH, Islam MS, Haque MA. Clinicopathological Study of Thyroglossal Cyst. Bangladesh Journal of Otorhinolaryngology. 2020 Dec 10;26(2):153-7.
- Delwar AH, Mazumder JA, Rashid MS, Mustafa MG, Hossain AB. Incidence and outcome of thyroglossal duct cyst. International Journal of Otorhinolaryngology and Head and Neck Surgery. 2020 Apr;6(4):616.
- Koirala K. Sistrunk's operation: ten years' experience in a teaching hospital. Nepal Journal Of Medical Sciences. 2020 Feb 15:5(1):20-7.
- Tymofieiev OO, Fesenko II, Cherniak OS, Zaritska VI. Features of Diagnostics, Clinical Course and Treatment of the Branchial Cleft Cysts. Diagnostics and treatment of oral and maxillofacial pathology. 2017(1,№ 1):15-31.
- Kim TH, Park YM, Cho JG, Woo JS. 2 Cases of Suprahyoid thyroglossal duct cyst with recurrent submental swelling. Korean Journal of Head & Neck Oncology. 2018;34(2):47-51.
- Verma R, Patro SK, Damodharan N, Sood A, Bal A. Papillary carcinoma thyroid in a thyroglossal cyst: A management dilemma. Acta Oto-Laryngologica Case Reports. 2017 Jan 1;2(1):5-10.
- Mortaja S, Sebeih H, Alobida NW, Al-Qahtani K. Large thyroglossal duct cyst: a case report. The American Journal of Case Reports. 2020;21:e919745-1.

- Isaacson G, Kaplon A, Tint D. Why Central Neck Dissection Works (and Fails) for Recurrent Thyroglossal Duct Remnants. Annals of Otology, Rhinology & Laryngology. 2019 Nov;128(11):1041-7.
- Patigaroo SA, Dar NH, Jallu AS, Ahmad R. Thyroglossal duct cysts: A clinicosurgical experience. Indian Journal of Otolaryngology and Head & Neck Surgery. 2017 Mar;69(1):102-7.
- Koirala K. Sistrunk's operation: ten years' experience in a teaching hospital. Nepal Journal Of Medical Sciences. 2020 Feb 15;5(1):20-7.
- Ogunkeyede SA, Ogundoyin OO. Management outcome of thyroglossal cyst in a tertiary health center in Southwest Nigeria. The Pan African Medical Journal. 2019;34.
- Von Bismarck S, Höllwarth ME. Thyroglossal duct cysts in paediatric patients: early operative intervention reduces rate of complications. Klinische Padiatrie. 2001 Sep 1;213(5):295-8.
- Simon LM, Magit AE. Impact of incision and drainage of infected thyroglossal duct cyst on recurrence after Sistrunk procedure. Archives of Otolaryngology–Head & Neck Surgery. 2012 Jan 16;138(1):20-4.
- Tamhane S, Gharib H. Thyroid nodule update on diagnosis and management. Clinical diabetes and endocrinology. 2016 Dec;2(1):1-0.
- Ostlie DJ, Burjonrappa SC, Snyder CL, Watts J, Murphy JP, Gittes GK, Andrews WA, Sharp RJ, Holcomb III GW. Thyroglossal duct infections and surgical outcomes. Journal of pediatric surgery. 2004 Mar 1;39(3):396-9.