



Recurrent thyroglossal duct cysts: a 15-year review of presentation, management and outcomes from a tertiary paediatric institution

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Background: Primary management of thyroglossal duct cysts (TGDC) is the Sistrunk procedure, which aims to completely excise the cyst and associated duct. Recurrences are attributed to variable histopathology and inadequate excision of the duct and its branching ductules. We present a review of the presentation and management of recurrent TGDC over a 15-year period to assess trends and outcomes.

Methods: Retrospective review of patients who underwent surgery for a TGDC at a tertiary paediatric hospital over a 15-year period. The following data was collected: age, sex, primary and subsequent presentations, histopathology, primary and subsequent operation reports and speciality of the primary operator.

Results: Sixteen patients had a recurrent TGDC over the 15-year period. Six (37%) were male and 10 (63%) were female, with a mean age of 5.3 ± 3.1 years. Initial management included Sistrunk procedure (11 cases, 68.75%), cystectomy (4 cases, 25%) and Schlange procedure (1 case, 6.25%). The mean time to recurrence was 20.7 ± 26.9 months. Management of recurrences were Sistrunk (5 cases), revision Sistrunk (9 cases) or conservative management (2 cases). Two patients had three operations, without cure.

Conclusions: Sistrunk procedure should be the primary management for TGDC. Patients who recurred after an initial limited surgical resection may be cured with subsequent Sistrunk procedure. Revision Sistrunk to manage recurrences following initial Sistrunk, had a high recurrence rate and two patients required two further operative interventions, both without cure. Research into the use of en bloc anterior neck dissection for the management of recurrent TGDC following Sistrunk in the paediatric population is suggested.

Keywords: Thyroglossal duct cyst (TGDC); thyroglossal duct sinus; recurrence; paediatric; management

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Introduction

Thyroglossal duct cysts (TGDC) arise from the incomplete closure of the thyroglossal tract, an integral structure in the embryological development of the thyroid gland. Thyroglossal tract remnants are reported to be present in 7% of the population, however not all develop a cyst (1,2).

TGDC are the most common congenital midline neck swelling in the paediatric population, with over half of all cases presenting before 20 years of age (3,4).

The management of TGDC has evolved through a greater understanding of the embryological and histological basis of the disease. Gold standard management is the Sistrunk procedure, which involves en bloc resection

of the cyst and the thyroglossal tract, with the superior excision encompassing the central aspect of the hyoid bone and continuing up to the foramen caecum, without breaching the oral mucosa (5). The recurrence rate after Sistrunk is between 3% and 10% (6-8), which is a vast improvement of the recurrence rate of more than 50% after performing an incision and drainage (I&D) (9). Recurrence is attributed to an inadequate initial resection along and/or variability in histological anatomy of the thyroglossal tract, with small ductules extending from the primary thyroglossal duct.

Typically, patients who recur present in a similar fashion to their primary presentation, however, there can be an increased incidence of cutaneous involvement (9). The majority of patients recur within the first postoperative year (10). The operative goal of recurrence surgery is to excise the remaining thyroglossal duct and associated ductules.

In this paper, we present a review of the presentation and management of recurrent TGDC over a 15-year period at a tertiary paediatric hospital to assess trends and outcomes.

Methods

Institutional board review was obtained. A retrospective chart review was performed to include all patients who presented to a tertiary paediatric hospital with a TGDC over a 15-year period. Patients were screened for recurrences of their TGDC through additional admissions to the tertiary hospital, clinic notes, primary care practitioner referrals and requests for admission sent from private consultant rooms.

Electronic and paper records were reviewed to obtain: age, sex, recurrence, primary presentation, primary management, presentation of recurrence/s, length of follow up, postoperative complications and further management. If patients were noted to have a recurrence they were sent a questionnaire with a single question, 'Have you/your child had another recurrence of a thyroglossal duct cyst or sinus since the last operation performed at The Children's Hospital at Westmead?'. If patients did not respond in 2 months, they were phoned.

Results

Two hundred and twenty-seven operations were performed for suspected TGDC or recurrent TGDC in paediatric patients over a 15-year period. Histopathology and

operation report review identified 191 (84%) cases of TGDC, 16 (7%) cases of recurrent TGDC, 13 (6%) cases of dermoid cysts and 1 (0.4%) each for epidermal cyst, neck abscess and lymph nodes. Patient notes were unable to be located for 4 subjects.

Recurrent TGDC

Demographics

Sixteen patients had a recurrence after initial management of their TGDC, of which 6 (38%) were male and 10 (62%) female. The patients had a mean age of 5.3 ± 3.1 years. *Table 1* outlines patient information.

Initial presentation and management

The initial TGDC presentation included non-infected cysts (9 cases, 56%), infected cyst (2, 12%), infected cyst and sinus (1, 6%), infected sinus (1, 6%), sinus (1, 6%) and the presentation was not documented in 2 cases (12%). The first surgical procedure of these patients included a Sistrunk procedure (11 cases, 68.75%), cystectomy (4 cases, 25%) and Schlange procedure (1 case, 6.25%). Four patients had an incision and drainage of an infected cyst, prior to initial cyst excision.

Presentation of recurrence

The mean time to recurrence was 20.7 ± 26.9 months. Patients presented with a non-infected cyst in 7 cases (44%), draining sinus in 7 cases (44%), cyst and sinus in 1 case (6%) and an infected cyst in 1 case (6%). Two patients had a third recurrence after operative intervention, 1 presented with a non-infected cyst and the other a draining sinus. Eight patients (50%) presented with a sinus at recurrence, of these, 3 (19%) had cutaneous involvement at initial presentation.

Management of recurrence

The management of recurrences was dependent on the primary operative intervention. Two operations were required for cure in 88% of patients. Eleven patients recurred after primary Sistrunk and of these, 9 were managed with revision Sistrunk and 2 were managed conservatively. The revision Sistrunk comprised of further dissection along the primary resection, with an extended excision of the remaining hyoid. Two patients (22%) had a further recurrence following revision Sistrunk, 1 was managed with a further revision Sistrunk, however recurred again, while the other had a cystectomy and also recurred.

Table 1 Presentation, management and outcomes for patients with a recurrent thyroglossal duct cyst

Case ID/Sex	Initial		1 st recurrence		2 nd recurrence		3 rd recurrence		Complication/ recurrence after last operative intervention	Incision and drainage prior to primary excision	Response to questionnaire	Total follow up (months)
	Presentation	Operation	Presentation	Operation	Presentation	Operation	Presentation	Operation				
1/F	Infected cyst	Sistrunk	Cyst	Revision Sistrunk						Yes	No recurrence	140
2/M	Cyst	Sistrunk	Cyst	Revision Sistrunk						No	No recurrence	180
3/F	Cyst	Sistrunk	Cyst	Revision Sistrunk						No	-	0.5
4/F	Cyst	Sistrunk	Infected cyst	Revision Sistrunk	Cyst	Cystectomy	Cyst	Monitored		No	-	1
5/M	Unknown	Cystectomy	Cyst	Sistrunk						No	-	0.5
6/M	Cyst	Sistrunk	Sinus	Revision Sistrunk	Sinus	Revision Sistrunk	Sinus	Monitored		Yes	-	3
7/M	Unknown	Cystectomy	Sinus	Sistrunk						No	No recurrence	180
8/M	Sinus	Sistrunk	Sinus	Monitored						Yes	No recurrence	36
9/F	Cyst	Sistrunk	Sinus	Revision Sistrunk						No	No recurrence	120
10/F	Infected sinus	Sistrunk	Cyst & sinus	Revision Sistrunk						No	-	6
11/F	Infected cyst & sinus	Sistrunk	Sinus	Revision Sistrunk						No	No recurrence	84
12/F	Cyst	Cystectomy	Sinus	Sistrunk						No	-	0.5
13/M	Infected cyst	Sistrunk	Sinus	Monitored						Yes	-	0.5
14/F	Cyst	Schlange	Cyst	Sistrunk						No	No recurrence	12
15/F	Cyst	Sistrunk	Cyst	Revision Sistrunk						No	-	0.5
16/F	Cyst	Cystectomy	Cyst	Sistrunk						No	No recurrence	12

To our knowledge, they were both subsequently managed conservatively. Four patients managed with a cystectomy recurred and in all cases, management with Sistrunk procedure provided cure. One patient recurred following Schlange procedure, which was successfully managed with Sistrunk procedure.

Response to questionnaire

Eight patients (50%) responded to the questionnaire, with no recurrences reported.

Follow up

Follow up ranged from 2 weeks to 15 years, with a mean of 48.5 ± 68.2 months.

Surgical speciality directing management

All patients who recurred had their initial intervention performed by a paediatric General Surgeon. Fifteen patients (94%) had their recurrence managed by a paediatric General Surgeon, while 1 patient (6%) was managed by a paediatric Otolaryngologist.

Discussion

TGDC recurrences are attributed to the incomplete excision of the thyroglossal tract and its branching ductules. The residual tract is present postoperatively due to variable histology or a limited surgical resection. Histologically, the thyroglossal duct can have multiple ductules extending from the main ventral duct which may not be detected intraoperatively, complicating the initial management and increasing the risk of recurrence due to residual thyroglossal tract. Historically, incision and drainage, cystectomy and Schlange procedure were performed to manage TGDC, however they are associated with high recurrence rates as they do not completely excise the thyroglossal tract, thus are no longer recommended treatment options. I&D prior to Sistrunk procedure has been associated with the need for multiple operations for cure (11). In our series, there were 4 patients who had an I&D prior to Sistrunk procedure. Three patients had no recurrence, however 1 patient has persistent disease despite 3 further operative interventions.

Failed surgical intervention alters the presentation of TGDC, with recurrences having a greater cutaneous involvement. In our series, 19% of initial TGDC presentation had a draining sinus, however with recurrent TGDC, the incidence rose to 50%. A previous review of

28 patients by Mickel *et al.* (9), found a similar increase in risk of cutaneous involvement following failed initial operative interventions. The majority of patients with a recurrence have been reported to present in the first postoperative year (10), however in our series, the majority presented in the second postoperative year. Younger children have a higher risk of recurrence, which has been suggested to be due to a conservative initial surgical approach, thus increasing the risk of residual thyroglossal tract (12,13).

The gold standard surgical management of TGDC is the Sistrunk procedure, as it offers the best chance of a complete excision of the thyroglossal tract and cyst. In our institution, there were 11 patients who had a recurrence following a primary Sistrunk procedure, 9 of whom underwent a revision Sistrunk, with the other 2 monitored. Seven (78%) of these patients had no further recurrence over a mean follow up of 21.8 ± 29.2 months. Two (22%) patients required further operative interventions without cure. Patients who recurred after being managed with a cystectomy or Schlange procedure (5 cases, 31%), had a subsequent Sistrunk procedure with no further recurrences. Thus, the Sistrunk procedure is a safe and effective operative intervention for patients who have a TGDC recurrence following inadequate initial resection.

The management of recurrent TGDC following Sistrunk procedure in children has been discussed, however papers are limited by patient numbers. The revision Sistrunk procedure, whereby the surgeon removes further tissue along the previous resection, has been well described in the literature. A systematic review by Ibrahim *et al.* (14) documented that the Sistrunk procedure was performed on 83 cases of recurrent TDGC in children, with 25 (30%) having further recurrences. In our series, 22% of patients managed with a revision Sistrunk had a further recurrence. Variations of an en block neck dissection have also been described in the literature, with an increased width of anterolateral dissection compared to the revision Sistrunk. Papers are limited by patient numbers and documentation of patient details, however the results are promising (9,15,16). A wide anterior neck dissection excising the central compartment between the lateral borders of the sternohyoid muscles has been performed in adults with recurrent TGDC, demonstrating good outcomes with minimal morbidity (17).

The subspecialty fellowship training of the primary operator has been reported to impact on the recurrence

rate, with fellowship trained otorhinolaryngologists having a lower recurrence rate than fellowship trained paediatric and plastic surgeons (18). In our review, all recurrences had their initial intervention performed by fellowship trained paediatric general surgeons and in similar fashion to Geller *et al.* (18), a proportion of these cases had an inadequate primary resection.

Our research is limited by small patient numbers, though this is expected given the low recurrence rate of TGDC after Sistrunk procedure and the relatively low incidence of the disease. Our patient numbers are in keeping with other published articles on the subject. We are also limited by the response of patients to the questionnaire, with half of the patients not contactable or declined participation. Given the long period of time between admission and research for this paper, many patients had changed address and contact details.

Conclusions

TGDC should be managed with Sistrunk procedure to minimise the risk of recurrence. The average age of patients in our series was 5.3 years old and they typically recurred within the second postoperative year. Patients had a higher risk of cutaneous involvement when a TGDC recurred. Sistrunk procedure should be considered in the management of recurrences after a limited initial surgical excision, as there were no reported recurrences in our series. Management of recurrence following initial Sistrunk is challenging, as revision Sistrunk had a high recurrence rate and 2 (22%) patients weren't cured despite 2 subsequent excisions. Further research into *en bloc* anterior neck dissections after failed Sistrunk in the paediatric population is required.

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Footnote

Conflicts of Interest: Both authors have completed the ICMJE uniform disclosure form (available at <http://dx.doi.org/10.21037/ajo.2018.05.02>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related

to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by the Sydney Children's Hospitals Network Human Research Ethics Committee, approval number LNR/14/SCHN/181. Informed consent was waived due to the retrospective nature of the study.

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