



Thyroglossal duct cyst and sinuses: A 20-year Los Angeles experience and lessons learned



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ARTICLE INFO

Article history:

Received 21 August 2013

Received in revised form 15 November 2013

Accepted 17 November 2013

Available online 25 November 2013

Keywords:

Sistrunk

Thyroglossal duct cyst

Thyroglossal duct sinus

Recurrence

Outpatient surgery

ABSTRACT

Background: First described in 1920 and later modified in 1928, the Sistrunk procedure substantially reduced the incidence of recurrence of midline neck cysts compared with a local excision or cystectomy. The purpose of this study was to determine if the rate of recurrence was influenced by performing either a 'classic' or a 'modified' Sistrunk procedure, if the recurrence rate was influenced by the physician's training, how successful we have been in managing patients with a recurrence? Finally, is outpatient surgery safe for Sistrunk procedures?

Methods: We performed a retrospective review of all patients with a thyroglossal duct remnant (TGDR) who were seen at the Children's Hospital Los Angeles (CHLA) from 1990 to 2010. The following data were collected: patient age, gender, presence or absence of a pre-operative infection, imaging studies, type of procedure performed, the attending surgeon's training background, inpatient or outpatient status, and complications.

Results: A total of 128 patients (61% male, 39% female) met the inclusion criteria. The age ranged from 2 months to 14 years (mean of 5.1 years). A total of 137 procedures were performed; 114 (83.2%) for primary and 23 (16.8%) for secondary disease. Complications included post-operative infection (10.9%), recurrence of disease (6.6%), undesirable scar (5.8%), and fistula (2.9%). Surgeons with fellowship-training in pediatric otolaryngology had a recurrence rate of 4.0% and surgeons with fellowship-training in pediatric surgery or pediatric plastic surgery had a recurrence rate of 30.1%. Twenty patients had a 'classic' Sistrunk (14.6%) and 117 (85%) had a 'modified' procedure. Patients were admitted after surgery in 78 cases (56.9%) and 59 patients (43.1%) had an outpatient (OPD) procedure.

Conclusions: There is no place for cystectomy in the treatment of TGDR. A 'modified' Sistrunk procedure is the procedure of choice in both primary and revision cases. Wide local excision of recurrences is required and a 'classic' Sistrunk should be considered. Specific training to gain an intimate knowledge of the anatomy in and around the larynx and experience with multiple cases reduces the incidence of recurrence. Outpatient surgery is safe and effective for selected patients who undergo a Sistrunk procedure.

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1. Introduction

In 1893, it was not Sistrunk but Schlange who first recommended excision of the center of the hyoid bone along with a midline cystic mass or a thyroglossal duct cyst (TGDC) which resulted in a reduction of the recurrence rate from as high as 50% for a cystectomy to about 20% [1]. In 1920, Sistrunk made his initial

contribution to this procedure with the recommendation to remove a one-eighth inch diameter core of tongue muscle superior to the hyoid at a 45 degree angle up to the foramen cecum to include mucosa [2]. Some other important parts of this procedure include removal of one-quarter inch of the center of the hyoid bone, closure of the cut ends of the hyoid bone, and placement of a drain. This is what we call a 'classic' Sistrunk procedure. Interestingly in this publication, there was no mention of a recurrence rate after performing this operation.

In 1928, Sistrunk modified the description of his original procedure by taking the dissection through the tongue base but not through the mucosa [3]. In fact, he specifically recommended

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placement of an index finger in the mouth over the area of the foramen cecum to insure that the surgeon not enter the oropharynx. This is what we call a 'modified' Sistrunk procedure. In this second publication, Sistrunk hinted at his experience with recurrence stating 'following such an operation practically 100 per cent of patients are cured'.

Over the last 20 years at our institution, we have seen significant variability of presentation of TGDCs and experienced the difficulty, occasionally, in removing them. This may be due to atypical presentations such as those reported recently by Sathish et al. and a history of prior infection [4,5].

The purpose of this study was to answer the following questions: have we routinely performed each step of the Sistrunk procedure? Is the rate of recurrence influenced by performing either a 'modified' or a 'classic' Sistrunk procedure? Is the recurrence rate influenced by the surgeon's training? How have we modified our own approach in dealing with recurrences and how successful have we been in managing patients with a recurrence? Finally, many surgeons in our group have regularly admitted patients post-operatively while one has most often performed this procedure as a same day surgery. Is it safe to perform a Sistrunk procedure as an outpatient surgery?

2. Methods

After obtaining institutional review board approval, a retrospective review of patients with thyroglossal duct cyst and sinuses or thyroglossal duct remnants (TGDR) seen at the Children's Hospital Los Angeles (CHLA) from 1990 to 2010 was completed. Inclusion criteria were documentation of the final pathology report as consistent with a TGDR and the presence of a complete pre-operative, operative and post-operative record. A total of 128 patients with 137 surgical cases met the inclusion criteria for this study. We collected the following data: patient age, gender, clinical presentation, presence or absence of a pre-operative infection, imaging studies, type of procedure performed, size and location of the cyst, sinus, or fistula, placement of a surgical drain, re-approximation of the hyoid bone, training background of the attending surgeon, inpatient or outpatient status with length of stay, post-operative infection, complications, and recurrence of disease.

3. Results

Of the 128 patients in the study, a total of 137 procedures were performed including treatment of both primary and secondary disease (Table 1). Primary disease, defined as a patient with a neck mass but without a history of any prior surgical procedure, was treated in 114 cases (83.2%). Secondary disease, defined as patient with a neck mass but with a history of a previous surgery, in 23 cases (16.8%). There were 9 cases of recurrence in the 137 procedures for an overall recurrence rate of 6.6% (Table 5). Of the 137 cases, an attending surgeon at CHLA with fellowship-training in pediatric otolaryngology performed 124 procedures with 5 cases

Table 1
General data.

Number of cases	137
Number of patients	128
Number with primary disease	114 (83.2%)
Number with secondary disease	23 (16.8%)
Males	78 (60.9%)
Females	50 (39.1%)
Mean age	5.1 years with range 2 months to 14 years
Number with pre-op infection	69 (50.4%)

Table 2
Imaging studies.

Imaging modality	Number of cases (n = 137)
Ultrasound	64 (46.7%)
None done	42 (30.7%)
CT scan	23 (16.8%)
MR scan	14 (10.2%)
Multiple modalities	12 (8.8%)
Thyroid scan	10 (7.3%)

of recurrence for a recurrence rate of 4.0%. Of the 137 cases, a surgeon with fellowship-training in either pediatric surgery or pediatric plastic surgery performed 13 procedures with 4 cases of recurrence for a recurrence rate of 30.1%. There was a male preponderance with 78 (60.9%) male and 50 (39.1%) female patients. The age at presentation ranged from 2 months to 14 years; with a mean of 5.1 years.

The most common symptom at presentation was a painless, midline, anterior neck mass. Movement of the mass with deglutition and fluctuation of the size of the mass were also common presenting signs. A history of previous infection was noted in 69 (50.4%) of cases. Dysphagia, cervical lymphadenopathy, drainage, infections requiring incision and drainage, and skin dimpling were less frequently seen. Imaging studies were obtained in many but not in all cases (Table 2).

The majority of patients had a 'modified' Sistrunk procedure (117 cases or 85%) compared with a 'classic' Sistrunk (20 cases or 14.6%) (Table 3). The two remaining ends of the hyoid bone were re-approximated in only 7 cases (5.1%). A drain was placed in 90 cases (65.7%). Patients were admitted after surgery in 78 cases (56.9%) while 59 patients (43.1%) had an outpatient (OPD) procedure. None of the OPD surgery patients had any airway compromise or were readmitted to the hospital emergently. Of those patients who were admitted immediately after surgery, the duration of hospitalization was 1–2 days (range 1–8 days).

Complications encountered included post-operative infection (10.9%), recurrence of disease (6.6%), undesirable scar (5.8%), and fistula (2.9%). Post-operative fistulas were seen in four patients. One of these patients had a total of four surgeries to treat this problem. All four of the patients who developed a fistula had undergone a 'classic' Sistrunk procedure. Other complications seen were drainage from surgical site, suture granuloma, and post-operative bleeding. One patient left the operating room intubated as a result of concern for airway compromise due to laryngeal edema (Table 4).

4. Discussion

We wanted to present our experience with the surgical treatment of TGDR's because of a number of issues raised recently and observations made over a 20-year period treating patients with this often challenging problem. The principal one was the fact that we have noted a significant number of referrals for patients

Table 3
Surgical procedure.

Procedure type	Number of cases (n = 137)
Modified Sistrunk	117 (85.4%)
Classic Sistrunk	20 (14.6%)
Hyoid bone ends reapprox.	7 (5.1%)
Drain placed	90 (65.7%)
Surgery as inpatient	78 (56.9%)
Surgery as outpatient	59 (43.1%)
Duration of hospitalization	1–2 days (range 1–8 days)

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