

Recurrent thyroglossal duct cysts: a clinical and pathologic analysis

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Abstract

Objective: To analyze, in detail, the clinical and pathological findings in a patient population that had undergone previous Sistrunk procedures for the removal of thyroglossal duct cysts, in order to identify any possible factors that could be related to recurrence after ‘definitive’ surgery. *Setting:* A large pediatric tertiary care center. *Methods:* A retrospective chart review was performed to include all patients treated at a single center for thyroglossal duct cysts with a Sistrunk procedure, between 1978 and 1992 inclusive. *Results:* A total of 108 consecutive patients were analyzed. We noted that the presence of recent preoperative infection of the cyst was the only statistically significant ($P < 0.05$) clinical difference noted between the successful and the unsuccessful surgical groups. However, pathological analysis revealed that there was a substantially greater number of multiple thyroglossal duct tracts ($P < 0.05$) noted in the group that failed a Sistrunk procedure. *Conclusions:* We feel that multiple thyroglossal tracts may play an etiologic role in some recurrent cysts. Thus, a wide conservative excision, including the middle two thirds of the hyoid bone, is necessary in order to include any multiple tracts in the resection. © 1998 Elsevier Science Ireland Ltd. All rights reserved.

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1. Introduction

The thyroid gland begins its embryological development at the level of the foramen caecum of the tongue. Subsequent descent of the bilobed

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thyroglossal diverticulum, will allow the thyroid gland to reach its normal position in the inferior neck area. The thyroglossal duct, connecting the thyroid gland with the foramen cecum, normally involutes beginning inferiorly. If this involution is incomplete, a remnant of the duct may persist in the upper neck area and contribute to the formation of a thyroglossal duct cyst.

Thyroglossal duct cysts are among the most common congenital cervical cysts noted in the pediatric population [1–3]. Classically, they present in young children as soft cystic midline swellings, that move superiorly with tongue protrusion. Historically, extraordinarily high recurrence rates noted with simple cyst excision, are thought to be related to the fact that persistent thyroglossal duct remnants were left in situ at the level of the hyoid bone and beyond [4]. In 1893, Schlange described a procedure that entailed the removal of the mid-portion of the hyoid bone in continuity with the main cyst [5]. In 1920, Sistrunk added excision of a block of tissue, between the hyoid bone and the foramen cecum, to the standard Schlange procedure, theoretically resulting in complete excision of the thyroglossal duct [6]. All procedures for removal of the thyroglossal duct that involve the removal of the central part of the hyoid bone, have come to bear Sistrunk's name. Although resulting in a substantial decrease in the postoperative recurrence rates, there remained a small, but definite failure rate following the Sistrunk operation. Most studies will note recurrence in approximately 7% (range 4–30%) of patients following a Sistrunk procedure [3,7,8].

In this article, we will review our substantial experience in the surgical treatment of thyroglossal duct cysts. We will review the clinical and pathologic findings in our patient population.

2. Materials and methods

We carried out a retrospective review of 108 consecutive pathologically confirmed cases of thyroglossal duct cysts that were surgically treated with a Sistrunk procedure at the Children's Hospital of Eastern Ontario in Ottawa, Canada, be-

tween 1978 and 1992. The patients were treated by various members of the Otolaryngology and Pediatric General Surgery services at this tertiary care center.

3. Results:

A total of 108 patients were operated on for thyroglossal duct cysts utilizing the standard Sistrunk procedure. Of these, 96 patients (88.9%) had no recurrence following Sistrunk, while 12 patients (11.1%) had one or more (range of 2–4, mean 2.4) further procedures for recurrent cystic disease of the thyroglossal duct (Table 1). No significant difference in recurrence rates could be identified in regards to a particular surgeon or surgical service performing the procedure.

Epidemiological data analysis revealed that thyroglossal duct cysts were most commonly noted in the midline (88.9% of cases). Furthermore, most affected patients were males (male/female = 1.6:1) with a mean age of 5.1 years. Only 17.7% of our patients had a preceding recent upper respiratory tract infection.

Intraoperative findings revealed the presence of a discernible tract leading up to the hyoid bone in 61.1% of patients (Table 2). There was intraoperative rupture of the cyst noted in 55.4% of patients.

Table 1
Demographic data comparison between patients having a successfully completed versus a failed Sistrunk procedure

	Successful Sistrunk	Failed Sistrunk
Sex (male/female)	60:36	7:5
Age at presentation (years)	4.9	6.7
Preceding upper respiratory infection (%)	17.4	20.0
Infected in 6 months preceding surgery (%)	44.8	75.0
Midline mass (%)	89.6	83.3
Elevation of cyst with tongue protrusion (%)	87.2	66.7

Table 2
Operative findings as noted at the time of Sistrunk procedure

	Successful Sistrunk	Failed Sistrunk
Tract seen during operation (%)	61.4	58.3
Cyst rupture during operation (%)	55.0	58.3
Sistrunk performed (%)	100	100
Base of tongue core-out performed (%)	80.0	75.0
Drain used postoperatively (%)	86.2	91.7

No significant statistical differences were noted in any of the above highlighted data between the patients who had failed a Sistrunk procedure and those who had a successful surgical intervention. Patients in the failed Sistrunk group did, however, have a statistically significant ($P < 0.05$; chi-square, single degree of freedom) increase in the incidence of preoperative infection of the cyst (75.0% versus 44.8% for successful surgical group).

There were surprising differences noted on pathological examination of the surgical specimens obtained from the two groups (Table 3). A single tract was noted to be present in only 16.7% of the recurrent cystic disease group, as compared to 46.7% for the successful Sistrunk group. Multi-

Table 3
Pathological analysis of surgical specimens analyzed following Sistrunk procedures

	Successful Sistrunk	Failed Sistrunk
Number of tracts visualized		
Zero	10.9	8.3
One	46.7	16.7
Multiple	42.4	75.0
Epithelial lining type		
Respiratory alone	30.2	33.3
Respiratory with squamous metaplasia	49.0	50.0
Nonspecific inflammation	20.8	16.7

ple tracts were more commonly noted in the recurrent cystic disease group ($P < 0.05$; chi-square, single degree of freedom). Thyroglossal duct cyst lining obtained from both groups revealed the predominance of respiratory epithelium, with or without squamous metaplasia and inflammation.

4. Discussion

Our institution's recurrence rate after Sistrunk procedure of 11.1% was within the range reported in the literature [3,7,8]. Recurrent disease after Sistrunk was noted to be associated with an increased rate of recent (previous 6 months) infection of the cyst. These increased recurrence rates following an infective episode, have also been reported by other investigators [8,9]. Notably, there appeared to be no increased risk of cyst recurrence if the cyst was ruptured intraoperatively. This differs from the findings of others [10]. Excepting the pathology findings, no other factors differentiated the successful from the non successful Sistrunk procedure groups.

Arborization of the thyroglossal duct at the level of the hyoid bone has been noted by a number of authors [9,11,12]. Multiple tracts occurred at a much higher rate in our patients that failed a Sistrunk procedure, as compared to the group that was cured by a Sistrunk procedure. Although the occurrence of multiple tracts is difficult to explain from an embryological standpoint, its occurrence may have clinical relevance. In order to encompass all of these tracts, we feel that it is necessary to resect the middle two thirds of the hyoid bone, or risk leaving some tracts in situ if a less conservative procedure is performed. Reports of multiple thyroglossal duct cysts occurring after an initial resection, likely represent the product of secretions accumulating within multiple residual tracts [13]. Any resulting recurrences are certainly more difficult to remove, as was noted by the frequently multiple excisions required to achieve cure, after a failed Sistrunk procedure. Based on histologic sections of the tongue, Horisawa et al. have, in addition, recommended the removal of a core from the base of

the tongue of approximately 5 mm in depth, in order to encompass the foramen cecum [14]. We found no significant differences between success or failure after a Sistrunk operation, in regards to the amount, if any, of tissue removed at the base of the tongue.

In summary, we have been able to identify that recurrence after Sistrunk procedures is more likely in patients who have had a history of recently infected thyroglossal duct cysts. Successful operative intervention seems to entail the removal of the cyst and a block of tissue encompassing the middle two thirds of the hyoid bone. Only with such a conservative approach will one be able to reliably include the full extent of the thyroglossal duct arborization. We feel that multiple tracts may possibly play an etiologic role in thyroglossal duct cyst recurrence. This point requires further clinical study.

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