Impact of Incision and Drainage of Infected Thyroglossal Duct Cyst on Recurrence After Sistrunk Procedure

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Objective: To determine whether incision and drainage of infected thyroglossal duct cysts (TGDCs) is associated with increased risk of recurrence after Sistrunk procedure when compared with antibiotic treatment alone.

Design: Retrospective case review.

Setting: Tertiary referral practice.

Patients: Patients treated for thyroglossal duct remnants in a tertiary care pediatric center from January 1, 2002, through December 31, 2008. Study subjects were identified using the diagnosis code for TGDC or the procedure code for excision of thyroglossal duct cyst or sinus.

Main Outcome Measures: The presence of infection, treatment of infected TGDCs, and relationship between infection, treatment of infection, and recurrence.

Results: We identified 120 patients from 2002 through 2008 who met the search criteria. The mean age at the time of presentation was 5.1 years; at the time of surgery, 5.4 years. More than half the patients (58.2%) were male, and the most common presentation was an asymptomatic midline neck mass. Forty-nine of our patients (40.8%) had a history of infection and 6 of these (12%) required incision and drainage. The overall recurrence rate was 10.8%. Ten of the patients with preoperative infection (20%) had a recurrence compared with 3 of the 71 patients (4%) without preoperative infection (P = .002). Of all patients with recurrences, only 1 had undergone incision and drainage.

Conclusions: This case series suggests that preoperative infection is associated with an increased recurrence rate. Incision and drainage of an infected TGDC may not increase the risk of postoperative recurrence. The results of this case series may assist in preoperative counseling and management of infected TGDC prior to definitive surgery.


In head and neck embryology, the thyroid gland descends into the neck as the lateral lingual swellings meet in the midline to form the tongue. During its migration, the middle portion of the gland remains attached to the foramen cecum at the base of the tongue by the thyroglossal duct. This connection tracks anterior to the hyoid bone and usually atrophies around the 10th week of gestation.1,2 Incomplete obliteration of the duct gives rise to thyroglossal duct cysts (TGDCs), which are the most common congenital neck mass with a 7% population prevalence.3-7

A thyroglossal duct cyst typically presents as a midline neck mass that fluctuates in size and has a tendency to become infected. Such a cyst also may contain viable thyroid tissue and can occur at any point along the thyroglossal tract.1,7 Less than half of these cysts develop into papillary thyroid cancer.8-10 The mainstay of treatment for TGDC is surgical excision using the Sistrunk procedure. The Sistrunk procedure was developed in 1920 and includes resection of the cyst, tract, midline portion of the hyoid bone, and cuff of the surrounding base of tongue musculature.11-13 Unfortunately, even with appropriate surgery, recurrence rates range from 1% to 30% in large series.2,3,14-25

Preoperative infection is the most common complication of TGDC, with rates ranging from 10% to 70%.2,3,14-25 Having 2 or more preoperative infections is associated with an elevated recurrence rate. Traditional thinking has been to treat preoperative infections with antibiotics alone. Needle aspiration may also be used in lieu of incision and drainage. This line of thinking is based on the idea that surgical drain-
age may increase scarring and obscure tissue planes, making eventual surgical extirpation more difficult.14-17 Our study reexamines the validity of this assumption and investigates not only the relationship between preoperative infection and recurrence but also the relationship between incision and drainage of infection and cyst recurrence.

METHODS

After obtaining approval from the Rady Children’s Hospital institutional review board, we queried the medical records of our otolaryngology practice from January 1, 2002, through December 31, 2008, for code 759.2 (TGDC) of the International Classification of Disease, Ninth Revision, and codes 60280 and 60281 (excision of TGDC/thyroglottal duct sinus and excision of recurrent TGDC/thyroglottal duct sinus, respectively) from the Current Procedural Terminology.26 Each record was individually reviewed, and the data extracted included age at presentation and treatment, sex, comorbid conditions, operation performed, presence of a malignant neoplasm, recurrence, complications, number of revision surgical procedures, presence of preoperative infection, and whether or not the infection had been treated with incision and drainage. Recurrence was defined as a persistent neck mass or a draining fistula requiring further excision. Postoperative infections requiring simple incision and drainage were not classified as recurrences. We excluded children who did not have TGDC as determined by results of the final pathological examination or who had received a primary operation at an outside facility. Statistical analysis using the Fisher exact test was used to compare the rates of recurrence with or without preoperative infection and with or without incision and drainage. Within the recurrence subgroup, the sample size was too small to perform inferential statistical analysis; therefore, we used descriptive statistics to analyze relationships.

RESULTS

Our query identified 120 patients who met the search criteria. The mean age at presentation was 5.1 years, and at the time of surgery, 5.4 years. Each child received a traditional Sistrunk procedure as their primary operation. More than half the patients (58.2%) were male, and the most common presentation was a painless neck mass (99.2%). Dysphagia (2.5%) and respiratory distress (<1%) were very rare. Six TGDCs were lingual, and only 1 patient had a malignant neoplasm, which was treated successfully with surgery. Forty-nine patients (40.8%) had a preoperative infection. Of these patients, 43 (88%) were treated with antibiotics alone, and 6 (12%) were treated with incision and drainage. The decision to perform incision and drainage was at the discretion of the attending pediatric otolaryngologist, and no specific criteria were used. In total, 13 patients (10.8%) had postoperative recurrences, 11 of whom presented within 1 year of the Sistrunk procedure. One presented at precisely 5 years and 1 presented 14 years after the initial procedure. Both patients had a history of preoperative infection, and neither patient had received incision and drainage. The data on these patients are listed in Table 1.

Of the 13 patients with recurrences, 10 (77%) had been treated for preoperative infection, and 3 (23%) had no history of infection. Ten of the 49 patients (20%) with preoperative infection had a recurrence compared with only 3 of the 71 patients (4%) without preoperative infection. A history of preoperative infection produced a statistically significantly higher rate of recurrence ($P=.007$, Fisher exact test). The relative risk for recurrence with a history of preoperative infection was 4.83 (95% CI, 1.40-16.65), and the odds ratio was 5.81 (95% CI, 1.51-22.30). These data are shown in Table 2.

Most recurrences appeared in patients with infection who were treated with antibiotics alone. Only 1 of the 10 patients (10%) with a history of preoperative infection who experienced a recurrence had received incision and drainage. Nine of the 10 patients (90%) with preoperative infection and recurrence were treated with antibiotics alone.

Only 6 of the 49 patients with preoperative infections received incision and drainage, and most of these patients (5 of 6 [83%]) did not have recurrences. Only 1 of the 6 patients (17%) treated with incision and drainage for preoperative infection experienced a recurrence.

### Table 1. Data on the 13 Children With Recurrence

<table>
<thead>
<tr>
<th>Agea</th>
<th>Preoperative Infection</th>
<th>Preoperative Incision and Drainage</th>
<th>Time From Initial Surgery to Recurrence, mo</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>No</td>
<td>No</td>
<td>9</td>
</tr>
<tr>
<td>2</td>
<td>No</td>
<td>No</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>Yes</td>
<td>Yes</td>
<td>4</td>
</tr>
<tr>
<td>2</td>
<td>Yes</td>
<td>Yes</td>
<td>11</td>
</tr>
<tr>
<td>4</td>
<td>Yes</td>
<td>No</td>
<td>5</td>
</tr>
<tr>
<td>4</td>
<td>Yes</td>
<td>No</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>Yes</td>
<td>No</td>
<td>3</td>
</tr>
<tr>
<td>6</td>
<td>Yes</td>
<td>No</td>
<td>72</td>
</tr>
<tr>
<td>7</td>
<td>Yes</td>
<td>Yes</td>
<td>9</td>
</tr>
<tr>
<td>8</td>
<td>Yes</td>
<td>No</td>
<td>10</td>
</tr>
<tr>
<td>13</td>
<td>Yes</td>
<td>No</td>
<td>12</td>
</tr>
<tr>
<td>6.5 wk</td>
<td>Yes</td>
<td>Yes</td>
<td>168</td>
</tr>
</tbody>
</table>

aUnless otherwise indicated, age is expressed in years.
Among all patients with preoperative infection, recurrence was similar in patients treated with antibiotics alone compared with those treated with incision and drainage. Of the 43 patients with preoperative infection who received antibiotics alone, 9 experienced recurrence (21%). Of the 6 patients treated with incision and drainage for infection, only 1 had a recurrence (17%). The Fisher exact test showed no statistical difference ($P > .99$) between rates of recurrence. This analysis is shown in Table 3 and the Figure.

**Table 2. Relationship Between Preoperative Infection and Recurrence**

<table>
<thead>
<tr>
<th>Patient Group</th>
<th>Recurrence $^a$</th>
<th>No Recurrence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Preoperative infection</td>
<td>10</td>
<td>39</td>
</tr>
<tr>
<td>No preoperative infection</td>
<td>3</td>
<td>68</td>
</tr>
</tbody>
</table>

$^a$Patients with preoperative infection had a higher rate of recurrence ($P = .007$, Fisher exact test; relative risk, 4.83; odds ratio, 5.81).

**Table 3. Relationship Between Incision and Drainage and Recurrence in Patients With a History of Preoperative Infection**

<table>
<thead>
<tr>
<th>Patient Group</th>
<th>Recurrence $^a$</th>
<th>No Recurrence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Antibiotics alone</td>
<td>9</td>
<td>34</td>
</tr>
<tr>
<td>Incision and drainage</td>
<td>1</td>
<td>5</td>
</tr>
</tbody>
</table>

$^a$We found no difference between groups with regard to rate of recurrence ($P > .99$, Fisher exact test).

Thyroglossal duct cyst is a common pediatric otolaryngologic problem, and many of these children require treatment for preoperative infection. Our patient demographics, rate of infection, and rate of recurrence are all consistent with the data found in previously reported large series.2,3,14-25

Our findings differ with regard to the proper management of preoperative infection. Traditional teaching has been to avoid incision and drainage and to rely on parenteral antibiotics and possible needle aspiration for refractory cases. The reason for this approach has been that incision and drainage will create scarring and obscure natural tissue planes, making dissection difficult and decreasing the likelihood of successful surgical extirpation. Prior studies examining this issue include Athow et al20 in 1989, Ostlie et al17 in 2004, and Marianowski et al15 in 2003. The studies by Athow et al20 and Marianowski et al15 found that the rate of recurrence was higher when incision and drainage was required than when it was not. In patients with preoperative infection who received and did not receive incision and drainage, Athow et al20 reported recurrence rates of 80% and 33%, respectively. Marianowski et al15 performed a statistical comparison and found similar results with recurrence rates of 31.25% when incision and drainage was required and 12% when it was not ($P < .05$). The findings by Ostlie et al17 differed. In their study, only 3 patients with infection had recurrences. Of these, 1 required incision and drainage and 2 did not.

Our study contradicts the findings of Athow et al20 and Marianowski et al15 with regard to the effect of incision and drainage on recurrence rate. Our patients had a 40.8% incidence of preoperative infection and a 10.8% recurrence rate. As with other studies, most of our 13 patients with recurrences (77%) had experienced preoperative infection, and this association was statistically significant ($P = .007$). However, unlike other studies, we found no statistical difference between incision and drainage and the use of antibiotics alone with regard to rate of recurrence (17% vs 21%; $P > .99$). In addition, patients with preoperative infections that recurred were much more likely to have been treated with antibiotics alone than with incision and drainage (90% vs 10%). Finally, patients who receive incision and drainage were more likely to have no recurrences (83% vs 17%). Unfortunately, our subgroups of patients with recurrences and those who received incision and drainage were too small to analyze with inferential statistics. However, we still found a strong trend suggesting that incision and drainage may not be associated with an elevated risk of recurrence. This finding warrants further investigation and suggests that the traditional paradigm of avoiding surgical treatment of infected TGDC may need to be reexamined. A randomized prospective trial would be best for such an investigation.

There are a number of possible explanations for our findings. First, incision and drainage may decrease the total duration of the inflammatory process. For example, a child treated with antibiotics alone may have erythema, warmth, and a persistent local inflammatory response for several days. However, the inflammatory process may be shortened by incision and drainage, resulting in more rapid healing and less scarring and tissue plane destruction. A second expla-
nation is that infections may follow 2 distinct courses. One course could be associated with diffuse inflammation throughout the cyst and surrounding tissues, whereas a second course produces a more localized process with a fluctuant cyst and very little surrounding inflammation. In this case, simple drainage of the cyst would treat the infection, and there would be little surrounding damage to obscure tissue planes. A third thought is that the extensive inflammation associated with incision and drainage and infections severe enough to warrant this therapy may actually result in scarring and destruction of the thyroglossal tract, similar to sclerotherapy obliterating a lymphatic malformation. In this case, although tissue planes are obscured, the damage to the cyst and tract by the disease process and treatment have worked to prevent recurrence instead of promoting it.

There are certain weaknesses with this study that must be addressed. First, although statistical significance was reached, we are still analyzing a small number of recurrences and infections. Only 6 patients underwent incision and drainage, and 1 of these patients had a recurrence. Owing to this sample size, descriptive statistics were used. With this analysis, we found that incision and drainage were not associated with a higher recurrence rate. However, if a larger patient set were analyzed, the results could be different. Given that the largest series of TGDC only includes 231 patients, a primary meta-analysis would likely be needed to accumulate enough patients for inferential statistics on incision and drainage among our patients. A further problem with the data set is that 2 of our recurrences in patients treated with antibiotics alone occurred at 5 and 14 years after the initial Sistrunk procedure. Both patients remained symptom free for many years and then presented with recurrent midline neck swelling. Results of pathological analysis from the revision procedure confirmed the diagnosis of recurrent TGDC. It is unclear why this happened, and such a large time span between the initial procedure and recurrence raises the possibility that these recurrences may not have had anything to do with the preoperative infections. Third, our search criteria were limited to patients whose medical records had the diagnosis code for TGDC or the procedure codes for the primary or revision Sistrunk procedure. Our search could have missed patients with codes for other diagnoses and treatments, such as dermoid cyst, who actually had a TGDC. As a result, we are not able to compare recurrence rates of simple cystectomy and the Sistrunk procedure. Also, it is possible that patients undergoing cystectomy may have had a higher incidence of preoperative incision and drainage and recurrence. Finally, our study examines only patients treated at a single institution by a single pediatric otolaryngology practice (Children’s Specialists of San Diego). Our experience and findings in this study are different from those at other large institutions. As such, a multi-institutional study including surgeons from varied training backgrounds is recommended to further examine our findings.

In summary, preoperative infection is common in TGDC and increases the risk of recurrence after the Sistrunk procedure. In our series, incision and drainage of these infections was not associated with an increased risk of recurrence when compared with treatment with antibiotics alone.

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Author Contributions: Both authors had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Simon and Magit. Acquisition of data: Simon and Magit. Analysis and interpretation of data: Simon and Magit. Drafting of the manuscript: Simon. Critical revision of the manuscript for important intellectual content: Simon and Magit. Statistical analysis: Simon and Magit. Administrative, technical, and material support: Simon and Magit. Study supervision: Magit.

Financial Disclosure: None reported.

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REFERENCES


