Preoperative evaluation of thyroglossal duct cysts: children versus adults—is there a difference?

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Abstract

BACKGROUND: Thyroglossal duct cysts (TGDCs) are common in children but also present in adults. This study evaluates the preoperative management and postoperative outcomes in patients with a resected TGDC.

METHODS: A retrospective analysis was performed on patients with a surgically treated TGDC. Clinicopathologic variables and treatment outcomes were obtained by chart review.

RESULTS: A total of 79 patients (44 pediatric and 35 adult) were identified. The majority of patients in both groups presented with a neck mass. Compared with children, adults were significantly more likely to undergo preoperative imaging and fine-needle aspiration biopsy. Malignancy was not identified in any patient on preoperative workup or postoperative pathology.

CONCLUSIONS: Adults with a TGDC are more likely to undergo preoperative imaging and biopsy. The infrequent occurrence of TGDC cancer or concurrent thyroid pathology suggests that the diagnosis of a TGDC may be made on clinical grounds in adult patients although ultrasound (± fine-needle aspiration biopsy) may be a useful supplementary modality.

The thyroglossal duct is an embryologic structure that represents a connection between the tongue at the foramen cecum and the developing thyroid gland. The duct normally involutes during the 8th to 10th week of gestation. The development of a thyroglossal duct cyst (TGDC) occurs if the duct remains persistently patent. TGDCs are the most commonly diagnosed neck mass in children; however, this condition is also prevalent in the adult population. These cysts are usually located inferior to the hyoid bone although they may occur anywhere between the base of the tongue and the superior aspect of the thyroid gland. Typically, the patient presents with a painless midline neck mass although occasionally the cyst may become infected and require a course of oral antibiotic therapy. Traditionally, the diagnosis of a TGDC is made on the basis of a thorough history and physical examination and is accomplished without the use of imaging or fine-needle aspiration biopsy (FNAB). TGDCs are generally excised using the Sistrunk procedure in which the cyst and its tract are resected along with the midportion of the hyoid bone, which ensures low recurrence rates.
This study was undertaken to examine the presentation and outcomes of TGDCs managed surgically at a single tertiary care institution with particular attention to differences in adult versus pediatric patients. Specifically, particular attention was given to the use of preoperative imaging and preoperative FNAB/cytology. Postoperative outcomes were also examined, with a specific focus on recurrence and final pathologic findings.

Materials and Methods

Study population

After obtaining institutional review board approval, the pathology database was used to identify patients with a histologically confirmed TGDC at University of Iowa Hospitals and Clinics, Iowa City, IA, between January 2000 and September 2011. Eighty-seven patients with TGDCs were identified. Patients with an incidental finding of a TGDC (noted during the management of a different disease process, n = 8) were excluded from the study population. A total of 79 patients formed the final study population.

Data acquisition

Patient charts were reviewed to obtain demographic information and clinicopathologic characteristics. Specifically, the initial presenting symptoms, use of preoperative imaging (including ultrasound, computed tomographic [CT] scanning, magnetic resonance imaging [MRI], and positron emission tomography), use of preoperative biopsy, and details of surgical treatment were noted. Follow-up data with respect to outcomes (including complications and recurrence) were also noted.

Statistical tests

Differences among patient groups were evaluated using the chi-square test. All statistical tests were performed using SPSS (IBM, New York, NY), and \( P < .05 \) was considered statistically significant.

Results

Demographics

A total of 79 patients were included in the final analysis. Pediatric patients were defined as any patient under the age of 18 years, and adult patients were defined as age 18 and older. A total of 44 pediatric patients and 35 adult patients were identified. Table 1 summarizes the demographic characteristics of these groups.

Presenting symptoms

Presenting symptoms and signs are shown in Table 2. The most frequent presenting symptom in both pediatric and adult groups was a neck mass (66% vs 71%, respectively). In the pediatric patients, neck swelling, neck infection, history of a TGDC, and dysphagia were also present. Adult patients presented with neck swelling (20%) and a prior history of a TGDC (6%), but none presented with neck infection or dysphagia. Three patients did not have documentation of primary presenting signs or symptoms.

Preoperative imaging

Overall, 45 (57%) patients underwent preoperative imaging studies. Within the pediatric group, 15 (34%) patients underwent imaging. Ten (67%) of these children underwent CT scanning only, 1 (67%) underwent ultrasonography only, and 2 (13%) underwent MRI only. One patient underwent both ultrasonography and CT scanning, and 1 patient underwent both MRI and CT imaging. Adults were significantly more likely to undergo preoperative imaging compared with children (86% vs 34.1%, \( P < .0001 \)). In the adult group, 30 (86%) patients underwent imaging. Table 3 summarizes all imaging undergone by the various groups of patients. Of these, 25 (83%) underwent CT imaging alone, 1 (3%) ultrasound alone, 2 (7%) MRI alone, and 2 (7%) both CT scanning and ultrasound. Two patients were found to have thyroid nodules while undergoing imaging for TGDCs.

Preoperative cytology

FNAB was performed in a total of 17 patients (22%) and was more frequently performed in adults versus children (43% vs 5%, \( P < .0001 \)). Nine (53%) patients who underwent FNAB were found to have “cystic contents” or provided a formal diagnosis of a “thyroglossal duct cyst.” The remaining 5 (29%) had nonspecific findings on FNAB, including adipocytes, histiocytes, and inflammatory cells. Two patients were found to have thyroid nodules on CT imaging or ultrasound. These nodules were biopsied and found to be benign. No patients undergoing imaging for a TGDC were found to have cancer in the thyroid gland itself.

Table 1  Patient demographics

<table>
<thead>
<tr>
<th></th>
<th>Adults</th>
<th>Children</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>N</td>
<td>35</td>
<td>44</td>
<td>79</td>
</tr>
<tr>
<td>Age, mean (range)</td>
<td>42.7 y (17.5–86.8 y)</td>
<td>6.1 y (5 wk–17.5 y)</td>
<td>22.3 y (5 wk–86.8 y)</td>
</tr>
<tr>
<td>M:F, %</td>
<td>63:27</td>
<td>55:45</td>
<td>58:42</td>
</tr>
</tbody>
</table>

F = female; M = male.
Operative procedure and final pathology

Most patients (98%) underwent a formal Sistrunk procedure. One pediatric patient underwent cyst excision alone without resection of the hyoid bone. On final pathology, none of the specimens were found to have a carcinoma within the cyst.

Outcomes

Follow-up data were available for 70 (87%) patients; the mean follow-up was 1.6 years (0 to 10.7 years). A total of 6 (11%) patients experienced a postoperative complication. The most frequent complication was dysphagia, which was experienced by 4 (5%) of the total study population. All of these patients were adults. One adult patient experienced a wound infection, and 1 pediatric patient experienced recurrent neck abscesses. Two pediatric patients experienced a postoperative seroma. One adult patient experienced chronic postoperative neck pain. One patient (1%) developed a recurrent TGDC. This was a pediatric patient who had undergone multiple prior procedures for infected TGDCs and floor of mouth abnormalities. The time to recurrence was 1 year from the Sistrunk procedure. This patient underwent revision of the initial cyst excision with core removal from the base of the tongue.

Comments

Because TGDC is the most common cause of a neck mass in children, it is considered primarily a pediatric problem. However, TGDCs may present in the adult population as well. Cadaveric studies have shown an incidence of asymptomatic TGDCs in 15% of adults. Interestingly, although the overall incidence of asymptomatic TGDCs is low, retrospective reviews have identified that between 56% and 69% of TGDC resections are performed in adults. In our study, 44% of patients were adults. Thus, it is necessary to maintain TGDCs in the differential diagnosis for adults presenting with a neck mass.

In children, a TGDC most often presents as a painless midline neck mass with less than 1% of TGDCs presenting outside of the midline. Ren et al2 showed that 66% of children presented with a neck mass with only 2.1% of children presenting with pain. This pattern was also shown by Shah et al3; 76% of pediatric patients with a TGDC presented with an asymptomatic neck mass. Our series confirmed this presentation in children, with 66% of pediatric patients presenting with a painless neck mass. Ostlie et al6 showed a preoperative infection rate of 18% in a cohort of 99 pediatric patients. In our series, 11.3% of children presented with neck infection, whereas no adults presented with infection.

The Sistrunk procedure, initially described in 1920, is well established as the definitive procedure for the treatment of TGDCs.7 This involves excision of the cyst itself, the tract of the thyroglossal duct, and the middle third portion of the hyoid bone. Although complication rates are low, potential complications vary from minor to major. Minor complications include a seroma, wound infection, stitch abscess, and wound dehiscence, whereas major complications include death, nerve damage, tracheotomy, abscess, and hematoma. TGDCs can also recur. A prospective study of 35 pediatric patients undergoing the Sistrunk procedure showed a minor complication rate of 29%. No patients in this study experienced any major complications or recurrence.8

Table 3 Preoperative imaging

<table>
<thead>
<tr>
<th>Imaging Type</th>
<th>Adults, n (%)</th>
<th>Children, n (%)</th>
<th>Total, n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Any imaging*</td>
<td>30 (86)</td>
<td>15 (34)</td>
<td>45 (57)</td>
</tr>
<tr>
<td>CT only*</td>
<td>25 (83)</td>
<td>10 (67)</td>
<td>35 (78)</td>
</tr>
<tr>
<td>Ultrasound only</td>
<td>1 (3)</td>
<td>1 (7)</td>
<td>2 (4)</td>
</tr>
<tr>
<td>Both CT and US</td>
<td>2 (7)</td>
<td>1 (7)</td>
<td>3 (7)</td>
</tr>
<tr>
<td>MRI only</td>
<td>2 (7)</td>
<td>2 (13)</td>
<td>4 (9)</td>
</tr>
<tr>
<td>Both CT and MRI</td>
<td>0 (0)</td>
<td>1 (7)</td>
<td>1 (2)</td>
</tr>
</tbody>
</table>

CT = computed tomography; MRI = magnetic resonance imaging; US = ultrasound.

*P < .05.
et al\textsuperscript{4} showed an overall recurrence rate of 5.9%. Interestingly, in their study, children who experienced recurrence underwent cyst excision alone, whereas all adults who experienced recurrence underwent a formal Sistrunk procedure.\textsuperscript{4} Factors reported to be associated with higher recurrence rates include an inaccurate initial diagnosis, infection, unusual presentation, lack of excision of base of tongue musculature, and presence of multiple thyroglossal duct tracts.\textsuperscript{5} The influence of preoperative infection on recurrence rates is controversial. Although Ostlie et al\textsuperscript{6} reported no correlation between preoperative infection and recurrence, Simon and Magit\textsuperscript{7} showed a significant correlation between preoperative infection and recurrence. Our findings are similar, endorsing a complication rate of 11% and a recurrence rate of 3%. The single recurrence in our series had undergone a formal Sistrunk operation but also had comorbid conditions including multiple floor of mouth fistulae. This patient had an unusual presentation and did not undergo excision of the base of the tongue musculature, factors that have both been suggested to correlate with recurrence.

In the adult patient, the most worrisome incidental finding in the workup and treatment of TGDCs is that of thyroid cancer within the cyst. Since the first report in 1911 by Brentano\textsuperscript{8} of TGDCa (thyroglossal duct carcinoma), approximately 200 cases have been reported in the literature. The incidence of TGDCa has been estimated to be .7% to 1.2%. Heshmati et al\textsuperscript{9} reported their experience at the Mayo Clinic with 741 patients treated for TGDCs with an incidence of TGDCa of .7%. In their study, 12 patients were diagnosed with TGDCa. Of those 12, 9 patients underwent thyroid exploration, and 5 were found to have concurrent intrathyroidal malignancies. Similarly, Plaza et al\textsuperscript{10} reported an incidence of 1.2% in a study of 511 patients who underwent TGDC excision. Only 1 patient in this study had intrathyroidal malignancy. These findings are similar to that of Patel et al\textsuperscript{11} in which no patient with TGDCa was found to have intrathyroidal malignancy, and the Sistrunk procedure was considered adequate resection for TGDCa.

Our data confirm that the incidence of thyroid cancer within lesions presumed to be primary TGDCs is low as is the incidental finding of other concurrent intrathyroidal malignancy. Two patients were found to have thyroid nodules on ultrasound, and these nodules were confirmed to be benign on FNAB. Furthermore, neither of these patients required thyroidectomy at the last follow-up (2.2 and 3.1 years). Despite these findings, it still appears that a majority of adult patients undergo preoperative imaging and FNAB as part of their workup. In light of this, it would be reasonable to suggest that adult patients with a high suspicion of TGDCs may be diagnosed clinically as well. This would be particularly true for individuals with typical clinical features (midline mass, which moves with protrusion of the tongue), no known risk factors for thyroid cancer, and no suspicious features on history and physical examination (eg, hoarseness, weight loss, or lymphadenopathy).

However, there are some limitations with regard to the current study aside from its retrospective nature. First, the true incidence rate of TGDCa may have been underestimated because of coding-related issues in the pathology database wherein TGDCa cancer would be coded as a carcinoma rather than a TGDC. In addition, our patient population is purely a surgical series, including only those patients who underwent resection of a primary TGDC. As such, we do not know the number of patients who presented with a midline mass but were found to have another pathology. Second, although a significant number of adults underwent imaging workup, most of these were CT scans, which are known to be less sensitive than ultrasound in identifying thyroid pathology. Therefore, it is likely that the rate of thyroid nodular disease was also underestimated in this study. Preoperative identification of nodules or cancers requiring intervention would be crucial to optimal initial surgical treatment.

In a survey from the United Kingdom, the majority of surgeons stated that they would use ultrasonography to investigate a suspected TGDC although only .17% of midline neck masses represented functioning thyroid tissue.\textsuperscript{12} We suggest that a presumed clinical diagnosis of a TGDC in an adult be supplemented by a neck ultrasound to evaluate both the presumed cyst and the remaining thyroid and nodal basins for additional pathology. The presence of any solid component or other suspicious findings should prompt fine needle aspiration and/or additional imaging for workup. This selective approach to investigations is also more likely to be cost-effective by allowing appropriate initial surgical therapy in patients who are found to have thyroid cancer. Larger, prospective, multicenter studies will be needed to establish the long-term validity of this approach.

References