Presentations and management of thyroglossal duct cyst in children versus adults: a review of 106 cases

Wenhao Ren, MD, Keqian Zhi, MD, DDS, Lu Zhao, DDS, and Ling Gao, DDS, Shaanxi, China
XI’AN JIAOTONG UNIVERSITY

Objective. The objective of this study was to determine the clinical presentations and management of thyroglossal duct cyst (TDC or thyroglossal tract remnant [TTR]) between children and adults and evaluate risk factors associated with recurrence after surgery for TDCs.

Study design. A retrospective study of all patients with TDCs managed in our department from January 1998 through April 2008 was performed. All records were reviewed for age and sex, sizes and locations of cysts, diagnostic methods, surgical management, recurrences, and complications. Differences between children and adults and risk factors associated with recurrence were evaluated.

Results. A total of 106 patients (47 children and 59 adults) were treated for TDC. Of the children, 57.4% were male and 42.6% were female, whereas 50.8% of the adults were male and 49.2% were female. There were no significant differences in sex in either group (P = .49). The average age was 7.0 ± 4.2 years in children and 36.0 ± 18.0 years in adults, which demonstrates a bimodal distribution. Adults were significantly more likely than children to present with a complaint other than mass or infection (P < .01), including pain, dysphagia, dysphonia, and fistula formation. There was no significant difference in frequency of location between the adults and children. In this article, 94.9% (56/59) of the adults and 87.2% (41/47) of the children underwent a Sistrunk operation, whereas the others underwent cyst excision. There were 2 recurrences among adults and 3 among children, all of whom presented with an infected neck mass and were treated with a second Sistrunk procedure. The recurrence rates and complications between children and adults were not significantly different.

Conclusions. The incidence of TDC was equal in males and females and had a bimodal distribution with similar incidence in children and adults. Adults were significantly more likely than children to present with a complaint other than mass or infection. The Sistrunk procedure is recommended as the main choice of treatment. Cyst infection may have a role in recurrence. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111:e1-e6)

Thyroglossal duct cysts (TDCs or thyroglossal tract remnant [TTR]) are the most common midline neck masses in childhood but can be found in adults.1,2 The standard surgical approach to TDC, encompassing removal of the midportion of the hyoid bone in continuity with the TDC and excision of a core of tissue between the hyoid bone and the foramen cecum, is often referred to as Sistrunk’s operation.3 Although the Sistrunk’s procedure is the standard treatment for TDCs, their management can still be a source of controversy in some instances.

Although TDCs are usually recognized in the pediatric population, and 50% to 60% of the lesions are diagnosed before age 20, the adult population still has this abnormality.1,4-6 There are few reports in the literature describing the differences in clinical presentations and outcomes of surgery between adults and children. We performed a retrospective study to find any differences in clinical presentations, diagnosis methods, treatment, and recurrence between children and adults. These results may be useful in predicting outcome and optimizing management of TDCs.

PATIENTS AND METHODS

A retrospective chart review was performed to evaluate patients with a diagnosis of TDCs in the Department of Oral and Maxillofacial Surgery, Xi’an Jiaotong University Stomatological Hospital, from January 1998 through April 2008.

A total of 116 patients with preoperative diagnosis of TDCs were identified in the medical records within this period. After reviewing surgical pathology reports, we
excluded 10 patients with misdiagnoses. The remaining 106 patients were divided into 2 groups: adults (≥18 years) and children (<18 years). The charts were reviewed, and the following information was recorded: age and sex, sizes and locations of cysts, diagnostic methods, surgical management, recurrences, and complications.

Continuous variables were presented as mean ± SD. The difference between children and adults was calculated using the $\chi^2$ test (or Fisher’s exact test) for categorical variables. The significance level for each hypothesis was .05. SPSS 13.0 for Windows (SPSS, Inc., Chicago, IL) was used for statistical analysis.

RESULTS

Age and sex

Of the 106 patients, 59 were adults and 47 were children; ages ranged from 6 months to 67 years. In the adult group, there were 30 males and 29 females, and in the pediatric population, there were 27 males and 20 females. There was no significant difference in sex between the children and adults ($P > .49$) and there were an approximately equal number of males (53.8%) and females (46.2%) in the overall population. The mean age of the children was 7.0 ± 4.2 years and the mean age of the adults was 36.0 ± 18.0 years. The average age at diagnosis was 28.0 ± 16.5 years in the overall population. There were 21 children (44.7%) who presented with TDCs before 5 years of age. In adults, TDCs were more frequently found between the ages of 21 and 30 years (24/59%, 40.7%) and between 41 and 50 years (18/59%, 30.5%). The occurrence in patients older than 60 years was rare (2/59%, 3.4%).

Presenting symptoms of thyroglossal duct cysts

The most common clinical presentation of TDCs is a neck mass, seen in 96 (90.5%) patients. Other presenting symptoms are shown in Table I. Adults were significantly more likely than children to present with a complaint other than mass or infection, including pain, dysphagia, dysphonia, and fistula formation: 54.2% (32/59) vs 23.4% (11/47).

Preoperative evaluation

The preoperative evaluation included physical examination, serum thyroid function tests, ultrasonography of the neck, computed tomography (CT), magnetic resonance imaging (MRI), radioisotope thyroid scanning, and fine-needle aspiration cytology (FNAC) (Table III). The most frequent preoperative study in adults was CT (54.2%), whereas it was ultrasonography (55.3%) in children. Neck CT scan and FNAC examinations were more frequently performed in the adult group than in children (CT: $P < .02$; FNAC: $P < .05$).

Surgery management

Ninety-seven patients were treated with Sistrunk’s procedure. Nine patients were treated with simple cyst
excision. All operations were performed by experienced surgeons (associate professor or above). The percentage of patients who received the Sistrunk operation was higher in adults than in children (94.9% versus 87.2%); however, surgical management was not significantly different between adults and children. All surgical specimens were sent for histopathologic examination. A suction drain was placed in 9 patients and a passive drain in 4. All patients underwent postoperative antibiotic therapy. Follow-up ranged from 2 to 48 months.

**Postoperative diagnosis, complication, and recurrence**

The preoperative diagnosis of TDC was histopathologically confirmed postoperatively in all 106 cases. No major complications were encountered. Minor complications were identified in 9 patients (8.5%), including local wound infections and seroma. This represents a total of 6 (10.2%) adults and 3 (6.4%) children with complications ($P > .37$). There were no cases of hypothyroidism secondary to the procedure. All complications were found in patients who underwent a Sistrunk procedure. Five (4.7%) patients had postoperative recurrence of TDCs: 3 in children after cyst excision and 2 in adults after the Sistrunk operation. All presented with an infected neck mass and were treated with a repeated Sistrunk procedure or extended Sistrunk procedure. All have had no recurrence. The recurrence rates were not statistically different between children and adults ($P > .39$). The rate of recurrence was not statistically different between cases with rupture of the cyst intraoperatively (1/12) and those cases without rupture (2/94) ($P > .22$). The rate of recurrence was also insignificant between cases with fistula or previous drainage of abscess (1/16) and those without fistula or previous drainage of abscess (4/99) ($P > .68$).

**DISCUSSION**

**Patient demographics**

TDCs are the most frequent congenital masses and cysts in the neck. Al-Khateeb and Al Zoubi reported that TDCs accounted for 53% of congenital neck masses in a series of 252 cases. In another series of 331 pediatric patients, TDCs accounted for 54.6% of all congenital cervical cysts. There was a controversy in age distribution of TDCs because some authors recorded the age at the time of onset in their series, whereas others recorded age at the time of initial diagnosis. In Allard’s meta-analysis, TDC incidence was found to be higher in children than in adults, whereas in other series, TDC was found to be more frequent in adults than in children. Our series similarly shows a more frequent occurrence in adults (55.7% versus 44.3%). The different results may be related to whether the age was recorded at the time of onset of symptoms or at the time of initial diagnosis.

Controversy about the sex distribution of TDCs also exists in the literature. Although in the largest series regarding pediatric congenital cervical cysts, Hsieh et al. observed a male predominance and other studies report a female predominance, most of the reviews reported an equal distribution of TDCs among males and females. In our series, we observed an equal distribution among males and females. These differences may be attributed to genetic and geographic differences.

**Clinical features**

The 2 main presentations of TDCs are that of a mass or infection, either as a single or a recurrent event. A TDC presents as a mobile, soft, painless mass if it is not infected. In infected cases, patients may have a painful neck mass, and develop fistula, dysphagia, cough, or respiratory obstruction. Allard reported that 32.6% of overall cases presented with fistulae in a series of 1534 cases. In our study, adults were significantly more likely than children to present with a complaint other than mass or infection ($P < .05$), including pain, dysphagia, dysphonia, and fistula formation. This result was similar to the rate in the study by Brousseau et al. Large TDCs at the base of the tongue may cause airway obstruction, which occurred equally in children and adults in this study.

According to knowledge of the embryogenesis and development of the tongue, thyroid gland, and hyoid bone, the TDC can be located anywhere from the foramen cecum in the tongue base to the suprasternal region. Most TDCs occur in close proximity to the hyoid bone. In a meta-analysis, Allard found that 2.1% of TDCs were lingual, 24.1% suprahyoid, 60.9% between the hyoid and thyroid cartilage, and 12.9% suprasternal. Although using a different terminology, Brousseau et al. found the locations to be similar to that presented by Allard in a series of 62 patients and there was no significant difference in the frequency of location between adults and children. Ahuja et al. reported 47.8% infrathyroid TDCs in children and 82.5% in adults. Shih-Tsang et al. also found a higher percentage of adults with cysts in the infrathyroid location (69.2% versus 37.5%). In our study, the overall distribution of location is similar to that presented by Allard and there was no significant difference in frequency of location between the adults and children.

**Preoperative evaluation**

Because of the close association with the hyoid bone and foramen cecum, movement of the cyst with swal-
lowing is often cited as a reliable diagnostic sign. A meticulous clinical history and physical examination are sufficient to make a correct preoperative diagnosis in most cases; however, the preoperative diagnosis could also emerge as a problem in TDCs with unusual locations. Opinions regarding preoperative evaluations in cases of presumed TDC varied in clinical practice.

The preoperative evaluation may consist of a thorough head and neck examination and it is important to identify the presence of functioning thyroid tissue, to confirm the diagnosis, and to detect malignant signs within the TDC. In our series, 6 main diagnostic methods were used in the preoperative evaluation for TDCs. These included ultrasonography, CT and MRI, FNAC, radioisotope thyroid scanning, and thyroid function test. CT and MRI play a supplementary role to more accurately delineate the anatomy of large cysts, and MRI may be used to define a residual fistulous tract in recurrent disease. Considering the relatively reasonable cost and useful diagnostic information, CT was the most common diagnostic tool for adults in this series. Ultrasonography was the most common test ordered in children, because it is readily available, inexpensive, noninvasive, and does not involve ionizing radiation or sedation, which is particularly important in children. It also offers valuable information for the identification of both the cyst and thyroid gland. If any solid element within the cyst is found, FNAC is a simple and reliable test that is useful in the differential diagnosis. However, with concern for possible injury, FNAC is not so popular for diagnosing TDCs in children. Although thyroglossal duct carcinoma is an uncommon complication of thyroglossal duct cyst, occurring in fewer than 1% of cases, the presence of large, atypical squamous cells or psammoma bodies in the FNAC test should suggest the presence of thyroglossal duct carcinoma.

There is controversy about the need for preoperative radioisotope thyroid scanning in patients with a presumed TDC. Advocates for routine preoperative radioisotope thyroid scanning are concerned with the possibility of ectopic thyroid as a midline neck mass. The excision of an ectopic thyroid gland that may be the only functioning thyroid tissue could lead to postoperative permanent hypothyroidism. The incidence of ectopic thyroid tissue, misdiagnosed as a TDC, is between 1% and 2%. However, like other centers, we are not in favor of routine preoperative isotope thyroid scanning. On the one hand, radioisotope thyroid scanning involves intravenous administration of radiopharmaceutical compounds, which should be minimized if possible, especially in the group of children; on the other hand, it seems superfluous if a normal thyroid gland can be identified on ultrasound.

### Surgical management

Before 1893, the treatment of TDCs was just a simple incision and drainage, and the recurrence was unacceptably high. Schlange was the first to describe the removal of the midportion of the hyoid bone in continuity with the TDC in 1893. Sistrunk added to Schlange’s procedure the excision of a block of tissue between the hyoid bone and the foramen cecum, and this remains the classic basic technique. The recurrence rate after the original Sistrunk procedure is about 3% to 4%. There are some reports about the modified Sistrunk’s procedure in the literature. Mondin et al. reported no recurrences in a group of 14 patients treated with a modified Sistrunk procedure, with resection of the cyst in continuity with the central portion of the hyoid bone, and skin island removal when fistulization was present. They doubted that core-out of tongue muscle was necessary. Bennett et al. reported no recurrences in a group of 20 patients treated without a core-out of tongue muscle; however, they supported the Sistrunk’s operation and did not state that core-out was unnecessary. Horisawa et al. reported a procedure similar to Sistrunk’s operation, except for a very limited core-out depth. The recurrence rate was no higher than that of the classical Sistrunk’s operation. In a survey regarding the treatment of TDCs in children in the United Kingdom, Brewis et al. reported that 19% of the surgeons excised only the TDC and the central portion of the hyoid bone. However, there was not a large series to compare the recurrence between Sistrunk’s procedure and the modified procedure. This issue would be best analyzed by a meta-analysis or a larger study. Some cases in our series were treated by local excision. This is performed only if the track is not identified at the posterior margin of the hyoid. We do favor the Sistrunk procedure as the surgical method of choice for the treatment of TDCs. A repeat Sistrunk procedure or extended Sistrunk procedure may be needed in cases of recurrence. In this series, considering the possible influence of normal craniofacial growth in children, the 3 recurrence cases in the children’s group were treated with repeated Sistrunk procedure, and the 2 adults with recurrence received the extended Sistrunk procedure. There was no recurrence.

### Complications

Possible major complications of TDC surgery are rare. They include recurrence, abscess or hematoma requiring surgical drainage, inadvertent entry into the airway, tracheotomy, nerve paralysis, hypothyroidism, and death. The minor complications may include seroma, local wound infection, stitch abscess, and wound dehiscence.
The Sistrunk procedure is known to be a relatively safe operation. Nevertheless, better knowledge of the complication rates could help us to improve the surgical technique and preoperative evaluation. Maddalozzo et al. report a 29% incidence of complications in 35 patients who were younger than 18 years, and another article reported a 12% incidence in adults. Brousseau et al. had a 24% complication rate in children and 7% in adults, including wound infection, seroma, and recurrence. Our results are consistent with these reports; even though the complication rate was higher in adults (9.4%) than in children (7.1%), the difference was not significant.

The recurrence rate after the Sistrunk procedure is reported to be about 3% to 6% in the most recent literature. In this study, the results were similar to the rate indicated in the literature and the recurrence rate was not significantly different between adults and children. Marianowski et al. reported that young age may be associated with an increase in recurrence. Türkyilmaz et al. reported the recurrence in pediatric patients was similar to the rate indicated in the literature (3.7%). Compared with the adult group, the recurrence rate of children was not significantly different in the literature. This is consistent with our results. Cyst infection may have a role in recurrences. Kaselas et al. found a significant relationship between the presence of preoperative inflammation, time of surgery, and recurrence of TDCs. They also demonstrated that no recurrences were found in patients with preoperative TDC inflammation who were initially treated with antibiotic administration and were operated on once the inflammation had settled. Ostlie et al. found postoperative infection was associated with a statistically significant incidence of recurrence. All of the 5 recurrences presented with preoperative cyst infection in this study; however, we did not find that postoperative infection was associated with an increased recurrence rate. There were no other related special features that could be identified, such as presentation with fistula, previous drainage of abscess, or rupture of the cyst intraoperatively.

Our data indicate that the incidence of TDC was equal in males and females and had a bimodal distribution with similar incidence in children and adults. Adults were significantly more likely than children to present with a complaint other than mass or infection, including pain, dysphagia, dysphonia, and fistula formation. CT was the most common diagnostic tool for adults, whereas ultrasonography was the most common test ordered in children in this series. Although a simple cyst excision is a choice in some cases, the Sistrunk procedure is recommended as the main choice of treatment. The recurrence rates and complications between children and adults were not significantly different. Cyst infection may have a role in recurrence.

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Reprint requests:
Keqian Zhi, MD, DDS
Department of Oral and Maxillofacial Surgery
Stomatological Hospital
College of Medicine
Xi’an Jiaotong University
98 Xiwu Road
Xi’an, Shaanxi 710004 China
zhikeqian@sina.com
zhikqian@mail.xjtu.edu.cn