



# Thyroglossal duct cysts: presentation and management in children versus adults

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**Summary Objectives:** To determine if the clinical presentation of thyroglossal duct cysts (TGDC) varies between children and adults and whether this knowledge helps optimize the surgical management. **Methods:** We retrospectively identified all patients with TGDC managed in our department between 1992 and 2002. We reviewed the patients' charts and recorded their gender, age at diagnosis, clinical presentation, radiologic imaging, surgical management, post-operative complications, and recurrence rate and compared the variables between the children and adults. **Results:** Twenty-one children and 41 adults were treated for TGDC. Of the children, 57% were male and 43% were female, whereas 49% of the adults were male and 51% were female ( $P = 0.53$ ). The average age was  $6 \pm 5$  years in children and  $45 \pm 16$  years in adults, which demonstrates a bimodal distribution. Forty-three percent of children and 42% of adults presented with an infected neck mass ( $P > 0.99$ ). Among our patients, 96% of the adults and 100% of the children underwent a Sistrunk operation. Four children developed a wound infection that resolved with antibiotics. One adult developed a haematoma and another developed a wound seroma. There was one recurrence among adults and one among children, both of whom were treated with a second Sistrunk procedure. **Conclusions:** There appears to be a bimodal distribution for age at presentation of TGDC. Since the differential diagnosis among adults is broader, the opportunity for misdiagnosis is greater. However, once the correct diagnosis is made, the surgical management and post-operative outcome between adults and children is the same.

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## 1. Introduction

A wide variety of neck masses are encountered in clinical practice (Table 1). Among these multiple

pathologies, thyroglossal duct cysts (TGDC) are the most common non-odontogenic cysts of the neck and the most common midline neck masses. The exact incidence of TGDC is unclear, but its incidence appears to be equal in both genders and is presumed to be higher in children than adults [1].

The clinical presentation of TGDC in children has been very well described [2]. Adult patients may have a more insidious presentation and the

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**Table 1** Differential diagnosis of a neck mass

Neck masses
Pyramidal lobe of the thyroid
Thyroid adenoma
Thyroid carcinoma
Thyroglossal duct cyst
Goiter
Aberrant thyroid tissue
Brachial cleft cyst
Dermoid cyst
Lymphadenopathy
Hemangioma
Lymphangioma
Salivary gland tumor
Laryngocele
Sebaceous cyst
Teratoma
Tuberculosis

differential diagnosis is wider, thus making the diagnosis of TGDC more difficult. There are no reports in the literature describing the differences in clinical presentation between adults and children. The knowledge of these differences could potentially be helpful in the management of both of these groups presenting with TGDC.

We performed a retrospective chart review of all the cases of TGDC managed in our department in the last 10 years to determine if the clinical presentation of TGDC varies between children and adults and whether this knowledge helps optimize the surgical management. For the first time, we contrast the presentation, treatment and complications associated with TGDC in adults and children.

## 2. Methods

### 2.1. Patients

We performed a retrospective chart review to identify patients treated for TGDC in the Department of Otolaryngology and Communicative Disorders at the Cleveland Clinic Foundation between 1992 and 2002. Using billing numbers, 70 charts were identified and 63 were available for review. After reviewing the 63 charts, we found that 62 contained the diagnosis of TGDC. Thus, 62 patients were included in the study. The charts were reviewed, and the following information was recorded: age at diagnosis, gender, presentation, family history, location of the mass, radiologic evaluation, pre-operative diagnosis, surgery performed, post-operative diagno-

sis, recurrence, treatment of recurrence and complications.

### 2.2. Statistical methods

Adult and pediatric groups were compared on the variables with two categories using Exact unconditional test, and on the variables with more than two categories using Fisher's exact test. The significance level for each hypothesis was 0.05. SAS software, Carey, NC, was used for all analyses.

## 3. Results

### 3.1. Patient demographics

Of the 62 patients, 41 were adults and 21 were children. There were an approximately equal number of males (52%) and females (48%) in the overall population. In the adult group, there were 20 males and 21 females, and in the pediatric population, there were 12 males and 9 females. The average age at diagnosis was  $32 \pm 22.7$  years in the overall population,  $45 \pm 16$  years in the adult group, and  $6 \pm 4.5$  years in the pediatric group. Presenting symptoms are shown in Table 2. One pediatric patient had a positive family history for TGDC; her mother had undergone a Sistrunk procedure as a child.

### 3.2. Pre-operative conditions

The pre-operative evaluation included physical exam, radiologic testing (i.e. neck CT, neck MRI, neck ultrasound, thyroid scan and barium swallow) and fine needle aspiration (Table 3). The location of the mass in relation to the hyoid bone, as well as to the midline in our cohort, is summarized in Table 4. Sixty cases were identified pre-operatively as TGDC. Two cases were identified post-operatively—one was initially diagnosed as a branchial cyst and the other was only identified as "neck mass." Six patients were known to have a recurrent TGDC, four were referred to us by another institution and two had been previously treated at our institution and experienced recurrent disease.

### 3.3. Surgical management

Fifty-nine cases were treated surgically with the technique originally described by Sistrunk [3]. Three (4%) of the adult cases and none of the pediatric cases were treated with simple excision. None of these patients had a recurrence. There were two recurrences among the patients who un-

**Table 2** Presenting symptoms of thyroglossal duct cysts

Presentation	Total no. (%) (n = 62)	Adult no. (%) (n = 41)	Children no. (%) (n = 21)
Mass	30 (48)	22 (54)	8 (38)
Recurrent mass	27 (44)	14 (34)	13 (34)
Infected mass	5 (8)	5 (12)	0
Recurrent infected mass	21 (34)	12 (29)	9 (43)
Otitis media	1 (2)	0	1 (5)
Pain	3 (5)	3 (7)	0
Sore throat	2 (3)	2 (5)	0
Dysphagia	9 (15)	8 (20)	1 (5)
Airway obstruction	2 (3)	1 (2)	1 (5)
Fistula	1 (2)	1 (2)	0
Globus	5 (8)	5 (12)	0
Hoarseness	3 (5)	2 (5)	0

**Table 3** Pre-operative evaluation

Work-up	Total no. (%) (n = 62)	Adult no. (%) (n = 41)	Children no. (%) (n = 21)
None	1 (2)	0	1 (5)
Ultrasound	20 (32)	10 (24)	10 (48)
CT Scan	35 (56)	25 (61)	10 (48)
MRI	2 (3)	1 (2)	1 (5)
Thyroid scan	3 (5)	2 (5)	1 (5)
Barium swallow	1 (2)	1 (2)	0
Fine needle aspiration	4 (7)	3 (7)	1 (5)

derwent their first Sistrunk at our institution: one among the adults and one among the children. Both patients underwent a second Sistrunk procedure. The pediatric patient required no further treatment, while the adult patient required a repeat Sistrunk with bilateral level 1A lymph node dissection. Four adult patients were referred to us for recurrent TGDC: one had multiple incision and drainage procedures as well as three excisions, one had two previous Sistrunk procedures and two had

one previous Sistrunk procedure. All were treated with a successful repeat Sistrunk procedure at our institution.

### 3.4. Post-operative diagnosis and complications

Post-operative diagnosis corroborated by pathology identified TGDC in 56 cases and recurrent TGDC in 6 cases. Post-operative complications were

**Table 4** Mass location

Location	Total no. (%) (n = 62)	Adult no. (%) (n = 41)	Children no. (%) (n = 21)
Relative to the midline			
Midline	22 (35)	17 (41)	5 (24)
Left	4 (6)	4 (10)	0
Right	4 (6)	1 (2)	3 (14)
Unspecified	32 (52)	19 (46)	13 (62)
Relative to the hyoid bone			
Submental	6 (10)	3 (7)	3 (14)
Suprahyoid	8 (13)	6 (15)	2 (10)
Over hyoid	30 (48)	17 (42)	13 (62)
Infrahyoid	17 (27)	14 (34)	3 (14)
Unspecified	1 (2)	1 (2)	0

encountered in eight patients (12.9%), including wound infection (5), seroma (1) or recurrence (2). This represents a total of three (7%) adult and five (24%) pediatric complications ( $P < 0.08$ ). There were no cases of hypothyroidism secondary to the procedure. All complications were found in patients that underwent a Sistrunk procedure.

## 4. Discussion

### 4.1. Patient demographics

In accord with the literature, we report an equal incidence of TGDC in males in females. However, contrary to previously described demographics [4–6] and commonly held beliefs, we report a high incidence of TGDC in adults. These findings are not altogether unusual because a number of other studies have also reported similar results, and these numbers may be directly influenced by local demographics [7–12]. Furthermore, close examination of the meta-analysis performed by Allard [1] reveals that about half of the cases present before age 20 and that there is a similar incidence between patients under 10 (31.5%) and above 30 (34.6%). Given these observations, we question whether the assumption of increased incidence of TGDC in childhood is truly substantiated. It may be more appropriate to claim that the most common age of presentation is the first decade of life, but there appears to be a bimodal distribution of TGDC as evidenced in our patient cohort.

### 4.2. Presenting symptoms of TGDC

Mass and infection, either as a single or recurrent event, are the two main presenting symptoms of TGDC. Other possible symptoms reported in the literature include: fistula, dysphagia, cough, respiratory obstruction and pain. There is little data as to the incidence of symptoms in patients with TGDC, but Josephson and Spencer [12] report that 65.7% of their patients presented with a mass or recurrent mass. In our study, a mass was present in 92% and infection in 42% of patients. Adults were significantly more likely than children to present with a complaint other than mass or infection ( $P < 0.024$ ), including pain, sore throat, dysphagia, hoarseness, globus and fistula formation. Airway obstruction occurred equally in children and adults. Otitis media appeared to be exclusive to children, although given the small number in our series is difficult to draw general conclusions. In this case,

TGDC was diagnosed during a thorough head and neck exam. This underlines the importance of assessing children for the presence of congenital abnormalities.

### 4.3. Genetics

Only one of our patients, a 6-year-old girl, had a family history of TGDC. There have been few reports of familial occurrence none of which described a clear pattern of inheritance [13–18]. However, a female predominance and some pattern of autosomal dominance have been observed [15]. It has also been suggested that the female predominance may be due to genetic imprinting, but there is no data supporting this theory at this time [15]. There may be a higher incidence of familial TGDC than that currently reported in the literature. Perhaps routine genetic testing could be useful in identifying patients with familial TGDC, which could lead to a better understanding of the etiologic factors relevant to this condition.

### 4.4. TGDC location

Basic thyroid, tongue and hyoid embryology is crucial to the understanding the possible anatomical positions of TGDC. The tongue has a mixed embryological origin. Its anterior section derives from the ectoderm and its posterior section from the endoderm. During the fourth week of development, the thyroid anlage is formed as an invagination of the endodermal cells in the tongue and descends as a bilobed diverticulum towards its mature position. The tract, followed by the thyroid, remains patent until the eighth week of development and is located ventrally to the developing hyoid bone and anterior to the thyroid membrane. Failure of the thyroglossal tract to involute can result in TGDC anywhere along its path. Furthermore, because the hyoid anlagen merge in the midline, the thyroglossal tract may become entrapped resulting in involvement of the periosteum or even the bone itself.

A meta-analysis by Allard [1] found that 2% of TGDC were intralingual, 24% suprahyoid, 60% between the hyoid and thyroid cartilage and 13% suprasternal. Although our data is presented using different terminology, the overall distribution of location is similar to that presented by Allard. There was no significant difference in TGDC frequency of location between the adults and children in our study. This observation suggests that TGDC pathophysiology must be similar in the two groups. More data regarding the location of TGDC in children and adults is needed to confirm this hypothesis.

#### 4.5. Pre-operative evaluation

The pre-operative evaluation of these patients may consist of a thorough head and neck exam with emphasis on identifying normally positioned thyroid tissue, and confirming the presence of normal thyroid tissue at the time of surgery. Pre-operative imaging such as ultrasonography, neck CT and MRI and radioisotope thyroid scanning can be used to identify the presence of normal thyroid tissue.

There is some controversy regarding the radiological investigation of TGDC. This issue is particularly important in children where inadvertent removal of ectopic thyroid gland that may be the only functioning thyroid tissue could lead to post-operative hypothyroidism and a life-long need for medication. The incidence of ectopic thyroid tissue, in the absence of a normally positioned thyroid, is reported to be between 1 and 2% [19].

According to a recent survey of pediatric surgical practices in the United Kingdom, ultrasound and radioisotope scans are the most common pre-operative imaging studies requested [20]. In our study, patients underwent either neck ultrasonography or CT. Despite the minimal use of thyroid radioisotope imaging none of the 62 patients developed hypothyroidism. We prefer ultrasound as the method of choice to determine the presence of normal thyroid tissue due to its low cost and availability. Nevertheless, complementary imaging may be used in selected cases based on the clinical presentation and pathological suspicion.

#### 4.6. Surgical management

Thyroglossal duct cysts have long been known to recur when treated with resection only. Wenglowksi [21] was the first to observe from his embryological studies that resection of the cyst together with the central portion of the hyoid and en bloc resection of the thyroglossal tract up to the foramen caecum would be necessary to prevent recurrence. It is based on these observations that Sistrunk designed his surgical technique. Other possible surgical approaches described include: Schlange procedure, marsupialization, shallow core out procedure and sclerotherapy [3,21–25].

One of our patients underwent three previous excisions before being referred to our institution, which underlines the importance of removing all elements of the tract at the time of surgery. Also, a thorough examination of the resected specimen may help prevent recurrences. Although some cases in our series were treated by local excision, we favor the Sistrunk procedure as the surgical method

of choice for the treatment of TGDC. Despite an adequate Sistrunk operation, some patients may develop recurrences. In our series, two patients originally treated at our institution with a Sistrunk operation developed a recurrence. In recalcitrant cases, the surgeon should consider further dissection of the neck to remove all tract remnants [26].

Our data set is too small to determine whether the recurrence rate was significantly different between the adults and children. Given the low recurrence rate after performing a Sistrunk procedure, this issue would be best analyzed by a meta-analysis or a larger study.

#### 4.7. Post-operative diagnosis

Post-operative diagnosis confirmed the pre-operative diagnosis of TGDC all patients included in this study. Pathological examination of all specimens that are removed is crucial to rule out TGDC cancer, particularly in older patients. Although TGDC cancer occurs in less than 1% [1] of TGDC cases, its presence would require a different management than simple TGDC. Although much controversy surrounds the appropriate management of TGDC cancer, recent studies have concluded that in most patients with a normal thyroid gland, the Sistrunk operation represents adequate treatment [27–29].

#### 4.8. Sistrunk complications

The Sistrunk procedure is associated with a diverse set of complications with an unclear incidence. Recent reports have found a 29% incidence of complications in patients who are younger than 18 years [30] and a 12% incidence in adults [12]. Our results are consistent with these reports. Even though the complication rate was lower in the adults (7%) than in the children (24%), the difference was not significant. However, this could be due to our small sample size. The Sistrunk procedure is known to be a relatively safe operation. Nevertheless, better knowledge of the complication rates could lead to improvement in surgical technique and peri-operative care of the patients.

### 5. Conclusion

In our study, the incidence of TGDC was equal in males and females and had a bimodal distribution with similar incidence in children and adults. The results of our study suggest that contrary to current opinion, the incidence of TGDC may not be greater in children, although the incidence is

generally higher in the first decade of life. However, these results may reflect biases due to referral patterns or may be just statistical noise in a small series. Although mass and infection were the most common presenting symptoms, the adults tended to present with a broader spectrum of symptoms, which may make the diagnosis of TGDC more difficult. The location of TGDC was similar in both children and adults, which suggests that the disease is caused by a single common pathophysiologic mechanism. We encourage the use of ultrasonography to identify the presence of normal thyroid tissue to prevent life-long hypothyroidism. Additional imaging studies may be helpful in selected cases where clinical presentation is confounding. Despite the variable complication rates between the children and adults, the Sistrunk procedure will most likely remain the treatment of choice for all patients with TGDC.

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