

# Thyroglossal cysts in a pediatric population: apparent differences from adult thyroglossal cysts

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**BACKGROUND AND OBJECTIVES:** The clinical presentation of thyroglossal cyst and its variation from adult thyroglossal cyst has not been well studied. This study is to determine if the clinical presentation of thyroglossal duct cysts (TGDC) varies between children and adults and whether this affects the optimal management of individual cases.

**DESIGN AND SETTING:** Retrospective study of all cases operated on for TGDC from February 2008 to November 2011 in a tertiary care teaching hospital.

**SUBJECTS AND METHODS:** The gender, age, clinical presentation, radiologic imaging, surgical management, post-operative complications, and recurrence rates between the children ( $\leq 18$  years) and adults ( $> 18$  years) were compared.

**RESULTS:** Of the 46 patients, 30 were adults and 16 pediatric; 46.5% of the adults and 74% of the children were females ( $P=.11$ ). The mean (SD) age in adults was 40.5 (16.0) years, while in children the mean (SD) age was 9.0 (1.4) years, suggesting a bimodal presentation. Hypothyroidism was present in 6.7% of the adults and 56.3% of pediatric cases ( $P=.0004$ ). Twenty percent of the adults had either an infected TGDC or fistulae, but none of the children had either infection ( $P=.0001$ ). In both adults and children the duration of disease was significantly shorter in females. In the pediatric group, males had a larger compared to females ( $P=.006$ ). The most common location of TGDC was the infrahyoid region. The Sistrunk procedure resulted in cure with no recurrences and complications in all.

**CONCLUSION:** TGDC is commoner in adults with no sex predilection. Children have a shorter duration of disease. Male children present with larger cysts. Hypothyroidism is more common in pediatric TGDC. Infected TGDC and fistulae are uncommon in children. The Sistrunk procedure is adequate for both groups.

The thyroid gland descends from the site of the foramen caecum in the base of the tongue to its final position between the fourth and seventh week of development. The thyroglossal tract disappears by the tenth week of gestation.<sup>1</sup> This tract, which extends from the base of the tongue to the isthmus of the thyroid, can be a site for thyroglossal cysts and ectopic thyroids.<sup>1</sup> Cadaveric studies have shown persistence of thyroglossal duct cysts (TGDC) remnants in 7% of an asymptomatic adult population.<sup>2</sup> TGDCs can occur anywhere along the tract of descent; however it is most commonly situated near the hyoid bone and the thyrohyoid membrane region (60%-65%).<sup>3</sup> Fifty to 60% of the TGDC are diagnosed in the pediatric popu-

lation, but some of them do present in adult life.<sup>4</sup> There are apparent differences in the proportions and types of clinical presentations of TGDC.<sup>5</sup> This study was done to determine if the clinical presentation, diagnostic methods, treatments and recurrences of TGDCs vary with the age of presentation (ie, children vs. adults).

## SUBJECTS AND METHODS

We retrospectively identified all patients with TGDC managed in our department between February 2008 and November 2011. Ethical clearance for the present study was obtained from the institutional ethical committee. Informed consent was obtained from the patients for surgery. All cases were operated on by the

**Table 1.** Demographics of the total group, pediatric and adult thyroglossal duct cysts (TGDC).

Variables	Total group (n=46)	Adult TGDC (n=30)	Pediatric TGDC (n=16)	Comparison of adult vs pediatric TGDC (P) <sup>a</sup>
<b>Sex</b>				
Male	43.5% (n=20)	53.3% (n=16)	25% (4)	.112
Female	56.5% (n=26)	46.5% (n=14)	74% (12)	
<b>Hypothyroidism (clinical/biochemical)</b>				
Yes	23.9% (n=11)	6.7% (n=02)	56.3% (n=9)	.0004
No	76.1% (n=35)	93.3% (n=28)	43.8% (n=7)	
<b>Clinical presentation</b>				
Neck mass	87% (n=40)	80% (n=24)	100% (n=16)	.0001
Infected mass	6.5% (n=3)	10% (n=3)	-	
Fistula	6.5% (n=3)	10% (n=3)	-	
<b>Location</b>				
Suprahyoid	21.7% (n=10)	20% (n=06)	25% (n=04)	NS
Over the hyoid	15.2% (n=7)	16.7% (n=05)	12.5% (n=2)	
Infrahyoid	63% (n=29)	63.3% (n=19)	62.5% (n=10)	

<sup>a</sup>Fischer exact test, NS: not significant

**Table 2.** Clinical characteristics of the total group, pediatric and adult TGDC.

Variables	Total group (n=46)	Adult TGDC (n=30)	Pediatric TGDC (n=16)	Comparison of adult vs pediatric TGDC (P) <sup>a</sup>
Duration of the cyst (months)	15.13 (12.37) (Range 2-60)	18.2 (13.83) (Range 2-60)	9.38 (5.89) (Range 3-21)	.02
Age (years)	29.52 (19.87) (Range: 8-70)	40.47 (15.98) (Range: 20-70)	9 (1.37) (Range: 8-11)	.0001
TSH (mIU/L)	6.21 (8.88) (Range 1.02-38)	2.6 (1.14) (Range 1.24-5.3)	12.99 (12.6) (Range: 1.02-38)	.0001
Size of the cyst (cms)	3.06 (0.77) (Range 1.8-5.1)	3.22 (0.8) Range (1.87-5.1)	2.78 (0.63) Range (1.8-3.8)	.06

<sup>a</sup>Independent sample t test

same surgeon in a single institution. We reviewed the patient charts and recorded gender, age at diagnosis, clinical presentation, radiologic imaging, surgical management, postoperative complications, and recurrence rates and compared the variables between the children ( $\leq 18$  years) and adults ( $> 18$  years). SPSS 13 statisti-

cal software package was used. (SPSS Inc., Chicago, IL, USA) Continuous variables were described as mean (standard deviation), categorical variables as numbers (percentages). For comparison between groups, independent samples t test and the Fisher exact test were used. Statistical significance was set at  $P < .05$ .

## RESULTS

Forty-six patients underwent surgery for TGDC during this period. There were 20 (43.5%) males and 26 (56.5%) females. Among these 30 patients were adults and 16 pediatric (34.78%) **Table 1** shows the demographics and clinical characteristics of the total group, with the pediatric and adult populations separately. Males and females were equally affected both in pediatric and adult patients, but the pediatric patients had a statistically significantly higher incidence of biochemical hypothyroidism ( $P = .0004$ ). In the pediatric group all presented with asymptomatic neck mass and none had fistulae or infection. This was different from the adults wherein some presented with infected cysts and fistulae ( $P = .0001$ ) (**Table 2**). The most common location of the TGDC was infrahyoid followed by a suprahyoid location in both the adult and pediatric groups (**Table 1**). TGDC in the pediatric age group was of shorter duration ( $P = .02$ ) and these patients had higher TSH values ( $P = .0001$ ). Comparison of the males with the females in the pediatric group revealed that the males had a larger TGDC ( $P = .0006$ ) at presentation, which was of shorter duration ( $P = .02$ ) (**Table 3**). In adults, even though the males had TGDC of longer duration ( $P = .02$ ), the size of the nodule was not statistically different between males and females ( $P = .11$ ) (**Table 4**). All suspected cases of TGDC underwent an ultrasound examination to look for the presence of a normal thyroid. Preoperative evaluation performed was either a combination of USG thyroid with Tc99 pertechnetate scan or an ultrasound of the thyroid alone. A combination of USG thyroid and Tc99 pertechnetate thyroid scan was undergone by 83.3% of the adults and 75% of the pediatric patients. Fine needle aspiration cytology (FNAC) is routinely performed in all cases of TGDC in the adult patients where we plan surgical intervention. In pediatric age group only 25% ( $n = 4$ ) underwent FNAC. All patients in the pediatric age group and the majority of the adults (90%) underwent the Sistrunk operation, whereas the patients with an infected mass underwent a drainage initially and the Sistrunk operation two months later. There were no recurrences in patients who underwent the Sistrunk operation. Histopathology did not reveal malignancy in the TGDC.

**DISCUSSION**

TGDC is one of the most common congenital neck masses. It moves on protrusion of the tongue. The differential diagnoses include dermoid cyst, lipoma, sebaceous cyst and others. Being a congenital anomaly it is generally believed that TGDC is common in children and adolescents, but in our series only approximately 35% belonged to this group. Brousseau et al<sup>5</sup> also noticed a higher incidence of TGDC in adults than in the pediatric age group. However, a meta-analysis by Allard et al<sup>6</sup> found a higher incidence of TGDC in the pediatric group (>50%).

No definite sex predilection was noticed in the pediatric and adult TGDC in our study. Males and females were equally affected. A similar observation was made in pediatric TGDC by others.<sup>6,7</sup> We observed that in the pediatric group the cyst was of a shorter duration and significantly more numbers were biochemically hypothyroid at presentation when compared to the adults (Table 2). The hypothyroidism had to be corrected before embarking on TGDC excision. In the pediatric group the males presented late ( $P=.002$ ) and had larger cysts ( $P=.006$ ). In adult TGDC the most common presentation was neck mass, infection or fistula, but the latter two features were absent in our pediatric group. Eighty percent of the adults and 100% of the pediatric group had presented with a neck mass in our series. Brousseau et al<sup>6</sup> observed that 88% of the adults and 72% of the pediatric group presented as a neck mass. They also reported infection in the TGDC in 43% of the children which was not observed by us. No fistula was noticed in pediatric group in their study.

Familial occurrence of TGDC has been reported. However, no such observation was made in this study.<sup>8</sup> In our study the most common location of TGDC was at the infrahyoid region in both the adult and pediatric groups (63.3% vs 62.5%). A similar observation was made by Allard et al (60% infrahyoid location). Preoperative evaluation of TGDC involves USG neck for assessing the presence of normal thyroid and also to assess the cyst. Tc99 pertechnetate is useful to diagnose the location of ectopic functional thyroid tissue especially if the thyroid gland is absent at its normal location. A combination of USG thyroid and Tc99 pertechnetate thyroid scan was undergone by 83.3% of the adults and 75% of the pediatric patients. A survey on the practice of pediatric surgeons of United Kingdom had revealed that they too preferred a combination of USG and Tc99 scans for evaluation of TGDC.<sup>9</sup> Some centers use either an USG or a CT, but USG, being cheaper and easily available, is widely preferred.<sup>6</sup>

The earliest operation for TGDC was simple inci-

**Table 3.** Clinical characteristics of males vs females in pediatric TGDC.

Variables	Males	Females	Comparison of males vs females TGDC ( $P^a$ )
Duration of the cyst (months)	16.5 (5.2)	7 (3.91)	.002
Age (years)	9.5 (1.73)	8.83 (1.27)	.41
TSH (mIU/L)	20.8 (20.32)	10.38 (8.65)	.16
Size of the cyst (cms)	5 (1.15)	2.85 (1.15)	.006

<sup>a</sup>Independent sample *t* test

**Table 4.** Clinical characteristics of males vs females in adult TGDC.

Variables	Males	Females	Comparison of males vs females TGDC ( $P^a$ )
Duration of the cyst (months)	23.38 (10.33)	12.29 (6.97)	.02
Age (years)	37.5 (10.6)	43.86 (20.38)	.28
TSH (mIU/L)	2.36 (0.92)	2.87 (1.33)	.23
Size of the cyst (cms)	4.14 (0.9)	3.58 (0.93)	.11

<sup>a</sup>Independent sample *t* test

sion and drainage; however it was associated with unacceptably high recurrence rates. Schlange proposed the removal of the central part of the hyoid bone along with the TGDC to prevent recurrences.<sup>10</sup> Sistrunk<sup>11</sup> added the removal of a block of tissue between the hyoid and foramen caecum to the Schlange procedure to decrease the recurrences. Several modifications were later described to the Sistrunk procedure. Mondin et al proposed removal of the skin island of the fistulae along with the tract. Several authors have reported low recurrences even if the tissue between the hyoid and foramen caecum is not removed.<sup>6</sup> Nineteen percent of the surgeons in United Kingdom were using only cyst excision.<sup>9</sup> However, it has to be stated that the Sistrunk procedure is based on the initial observations of Wengłowski et al on the embryological development of the TGDC.<sup>6</sup> We have had no recurrences in cases where the Sistrunk procedure was used. Malignancy has been reported in TGDC, but in our cases we did not find any. The incidence of malignancy has been 1%.<sup>7</sup> Both papillary thyroid cancer and squamous cell carcinoma (SCC) have been described.<sup>12</sup> Both of these have been described in adults as well as pediatric patients. Even though SCC is dealt with by wide local excision of the cyst while performing the Sistrunk procedure, the management of papillary carcinoma in TGDC is controversial. Many have suggested that the Sistrunk

operation is an adequate procedure,<sup>6</sup> but some authors do a total thyroidectomy<sup>13</sup> so as to ensure proper follow up with thyroglobulin estimation and radioiodine scans. Rarely Hashimoto's thyroiditis has also been observed in patients within ectopic thyroid tissue within the TGDC.<sup>14</sup>

To conclude TGDC can present in both adults and children. There is no predilection for any particular sex.

TGDC is of a shorter duration in children than adults especially in female children. The cyst is larger in size in male children when compared to the females. Pediatric TGDC are commonly associated with biochemical hypothyroidism. Infected TGDC and fistulae are commonly seen in adults. The surgical procedure in both groups is the Sistrunk procedure. The Sistrunk procedure is adequate for TGDC both in children and adults.

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