



## Original Article

# PRESENTATION OF THYROGLOSSAL CYST IN CHILDREN

SNF RUMI<sup>1</sup>, SM AHMAD<sup>2</sup>, S RAHMAN<sup>3</sup>, AH TABLU<sup>4</sup>

### Abstract

**Background:** Thyroglossal duct cyst a developmental anomaly present as a congenital cervical masses of neck in children.

**Objectives:** To observe its deferent presentation and evaluate among the children.

**Materials and methods:** This observational cross section study conducted among patients of eighteen years of age present with thyroglossal duct cyst between 2007 to 2012 in the department of ENT and Head-Neck surgery, Dhaka Medical College Hospital, Dhaka. All patients were operated by Sistrunk operation under (excision of total cyst and removal median portion hyoid bone) under general anesthesia. All specimens were histo-pathologically confirmed.

**Results:** Among 24 children with thyroglossal cyst, 15 boys and 9 girls with male female ratio 1.67:1, age ranged from 4 years to 18 years (mean 9.46 std  $\pm$ 4.27). Male (mean 7.53  $\pm$ 4.01 years) child are younger than female (mean 12.67  $\pm$ 2.39 years) child. 66.67% male children were below 10 years of age and 88.89% female children were over 10 years of age. According to the presentation site 17 (70.83%) cases were juxtra hyoid, 4 (16.67%) were suprahyoid and 3 (12.50%) were infrahyoid. 23 (95.83%) were present as midline swelling, only 1 (4.17%) was present as left lateral infrahyoid swelling. 4 (16.67%) patient were attended as thyroglossal fistulae with

history of intervention. 4 (16.67%) patient were developed recurrence followed by surgical resection within two years.

**Conclusion:** Thyroglossal duct cyst present as an asymptomatic midline neck mass around the hyoid region in children. Complete resection along with median portion hyoid bone prevents recurrence.

**Key words:** Thyroglossal duct cyst, developmental neck mass, midline neck swelling.

### Introduction

A Thyroglossal duct cyst is a developmental anomaly, which is cystic dilatation of the persistence epithelial remnants of the thyroglossal duct tract. Thyroglossal duct cyst (TDC) is the most common benign developmental lesion of the neck, constitute 70% of all the congenital cervical masses<sup>1</sup>. They comprise approximately one-third of congenital neck masses in children<sup>2</sup>, second most common of all childhood cervical neck masses<sup>3</sup>.

It develops from the persistence of thyroglossal tract during the development of the hyoid bone and subsequent embryogenesis and descent of the thyroid gland and failure of complete obliteration of the duct<sup>4</sup>.

During the fourth week of development the thyroid enlarge arises as an invagination of endoderm in the floor of pharynx between the tuberculum impar and posterior third of the tongue. During embryogenesis thyroid primordium migrate caudally with the descent of great vessels. It passes either anterior, posterior or through the hyoid bone in mid line of the neck<sup>5</sup>. During its migration the gland remains connected to the tongue by a narrow canal, the thyroglossal tract. This tract normally atrophies and disappears between fifth to tenth weeks, though the caudal end often remains as pyramidal lobe among one third of people. No

1. Dr. Shaikh Nurul Fattah Rumi, Associate Professor, Department of ENT & Head-Neck Surgery, Dhaka Medical College, Dhaka.
2. Dr. Shaikh Muniruddin Ahmad, Junior Consultant (ENT), Upazila Health Complex, Kapasia, Gazipur.
3. Dr. Shahnaz Rahman, Senior Medical Officer, Department of Gynae & Obstetric, BIRDEM Hospital 2, Segunbagicha, Dhaka.
4. Prof. Dr. Abdul Hanif Tablu, Department of Pediatric Surgery, Dhaka Medical College, Dhaka.

**Correspondence to:** Dr. Shaikh Nurul Fattah Rumi, Associate Professor, Department of ENT & Head-Neck Surgery, Dhaka Medical College, Dhaka. E-mail: rumi17dr@gmail.com

natural internal opening of thyroglossal duct has been demonstrated at the level of foramen cecum as the tongue and foramen cecum forms after the complete descent of the thyroglossal duct. Rarely a tract could be found at the level of foramen cecum.

Portions of this epithelial tract and remnants of thyroid tissue may persist, due to failure of the complete obliteration of the tract. It may develop anywhere along the course of the duct remnant, from the base of the tongue to the suprasternal region. The commonest site of this is just above or just below the hyoid bone<sup>6</sup>.

The Thyroglossal duct cyst may be formed as a result of undergoing cystic degeneration of epithelial remnants of the persistent tract due to recurrent throat inflammation and or a blocked thyroglossal duct expands as a result the of retention of secretion<sup>7</sup> as the continuous mucous production from the glands found in the duct<sup>8</sup>.

Cysts located near the foramen caecum are lined by pseudo stratified ciliated columnar with some squamous epithelium, whereas cysts located near the thyroid gland are lined by cells similar to thyroidal acinar epithelium<sup>9</sup>. Functional thyroid tissue within the TDC has been described and more than half contain normal thyroid tissue in their walls.<sup>10</sup>

Any part of the tract can persist causing a sinus, fistulae or cyst. Formation of the cyst is likely. Most fistulae and sinuses are acquired following rupture or incision of infected thyroglossal cyst. Occasionally, a sinus tract is present in the midline without a visible cyst<sup>11</sup>.

It may be found in patients of any age<sup>12</sup> with equal male-to-female incidence<sup>13</sup>. The Thyroglossal duct cyst (TDC) is a developmental anomaly, but it does not appear as a congenital lesion<sup>14</sup>. Up to 50% thyroglossal cysts are not diagnosed until adult life. The tract can lie dormant for years or even decades until some kind of stimulus leads to cystic dilation. It is more common in children<sup>15</sup>. Typically it appears in a person over the 5 years of life<sup>16</sup>. 60% of lesions are diagnosed before the age of 20 with over 30% presenting before ten years of age.<sup>17, 18</sup>

It is mostly asymptomatic. Some patients will have neck or throat pain or dysphagia due to the size of this mass or due to infection.

Diagnosis is usually clinical, history and physical examination are required for the correct diagnosis. Upwards movement during swallowing or on protrusion

of the tongue (because of its attachment to the tongue via the tract of thyroid descent) is its cardinal sign.

Ultrasonography will help to confirm the diagnosis and identify the presence of normal thyroid gland in neck<sup>19</sup>. Commonly this cyst could contain thyroid tissue. Thyroid scans with I<sup>131</sup> is needed when normal thyroid tissue is not found or all patients with suprahyoid masses<sup>20</sup>. Rarely this could be the only functioning thyroid gland tissue.

Thyroglossal cysts are treated surgically. Although generally benign the cyst will be removed if the patient exhibits difficulty in breathing or swallowing, or if the cyst is infected. Even if these symptoms are not present the cyst may be removed to eliminate the chance of infection or development of a carcinoma<sup>21</sup>, or for cosmetic reasons if there is unsightly protrusion from the neck.

The surgical resection involves excision of the cyst along with its tract and concomitant removal of the central portion (midsection) of the hyoid bone (to ensure complete removal of the tract) for avoid recurrence. It is called Sistrunk procedure. Wenglowski who suggested that along with the body of hyoid bone a core of tissue between the hyoid bone and the tongue should also be removed to reduce the incidence of recurrence.

The purpose of this study was to review the cases and describe the clinical presentations and site distribution of the thyroglossal cyst in our experience. Another aim is to ensure the absence of the recurrences by correct removal of the cyst.

## Materials and Methods

This cross sectional observational descriptive study was conducted at the Department of ENT and Head-Neck Surgery, Dhaka Medical College Hospital, Dhaka. from January 2007 to December 2012. A total of twenty four patients of ages below 18 years and both sexes who were operated during the study duration were included in the study. Patients above 18 years were excluded from the study. A detailed history and thorough physical examination was conducted. Ultrasound studies were done in all cases to confirm the cystic nature of the lesion and the presence of normal thyroid gland at its normal site. FNAC was also done preoperatively to exclude other pathology and to study the type of pathology histopathology also done. Thyroid scan was done to confirm the lesion are the only thyroid tissue or not and normal thyroid gland present or not.

A semi structured proforma was made to collect all the necessary information of the patient available. Management was mainly surgical and Sistrunk operation was done in all cases. Cyst was removed through a horizontal incision made at the inferior border of the mass along with body of the anomalies a core of suprahyoid muscles was removed. This was done to prevent any recurrence. Follow up was done at one, two, four week and six months interval for any complications and recurrence.

**Results**

Among 24 children (aged before 18 years) of thyroglossal cyst, 15 boys and 9 girls with male female ratio 1.67:1, age ranged from 4 years to 18 years (mean 9.46 std ±4.27). Male (mean 7.53±4.01 years) child are younger than female (mean 12.67±2.39 years) child. 66.67% male children were below 10 years of age and 88.89% female children were over 10 years of age.

According to the presentation site 17(70.83%) cases were juxtra hyoid, 4(16.67%) were suprahyoid and 3 (12.50%) were infrahyoid. 23 (95.83%) were present as midline swelling, only 1(4.17%) was present as left lateral infrahyoid swelling. 4(16.67%) patient were attended as thyroglossal fistulae with history of intervention. 4(16.67%) patient were developed recurrence followed by surgical resection within two years. Histopathologically one cyst was reported as papillary carcinoma other were reported benign. In two patient (8.34%) it was the only thyroid tissue of the neck.

**Table-I**  
*Age and Sex distribution of the children with Thyroglossal cyst (n=24)*

Age	Male	%	Female	%	Total	%
<5yrs	3	20.0	0	0.00	3	12.5
5-10yrs	7	46.6	1	11.1	8	33.3
>10yrs	5	33.3	8	88.9	13	54.2
Total	15	100.0	9	100.0	24	100.0

Chi-square= 7.138 with 2 degrees of freedom. (P = 0.028)

**Table-II**  
*Age of the children in relation to their sex with Thyroglossal cyst (n=24)*

Sex	Number %	Mean	Std
Male	15 (62.5%)	7.53	4.02
Female	9 (37.5%)	12.67	2.40
Total	24 (100%)	9.46	4.27

t = -3.464 with 22 degrees of freedom. (P = 0.002)

**Table-III**  
*Presentation of the Thyroglossal Duct Cyst (n=24)*

Presentation	No	%
Neck mass (midline)	23	95.83
Discharging sinus	1	4.17
Globus	6	25.00
Dysphagia	2	8.33
Sore throat	2	8.33
Pain	3	12.50
Hoarseness	1	4.17
Others	1	4.17
Total	24	100

Most of the patient (95.83%) was presented as an asymptomatic midline neck mass which gradually increasing in size. Six (25%) children had globus like sensation and three (12.5%) children had pain due to infection. Only 8.33% children had the complaint of dysphagia or sore throat.

**Table-VI**  
*Sonographic features of the Thyroglossal Duct Cyst (n=24)*

Sonographic Features	No	%
Echogenicity		
Anechoic	8	33.33
Hypoechoic (homogenous)	8	33.33
Hyperechoic (complex)	4	16.67
Hyperechoic pseudosolid	3	12.50
Hyperechoic Solid	1	4.17
Posterior enhancement		
Well defined	21	87.50
Subtle	3	12.50
Cyst Wall		
Thin	15	62.50
Thick	5	20.83
Imperceptible	4	16.67
Total	24	100.0
Size (mm)	Mean	SD
Range 10 – 30mm	19.04mm	4.66mm

**Table-V**  
*Presentation of the Thyroglossal Duct Cyst according to their sites (n=24)*

Character	Supra hyoid	%	Juxtrahyoid	%	Infrahyoid	%	Total	%	
Side	Midline	4	100	17	100	2	66.7	23	95.8
	Left	0	0	0	0.0	1	33.3	1	4.2
	Right	0	0	0	0.0	0	0.0	0	0.0
Age	<5yr	2	50	1	5.9	0	0.0	3	12.5
	5-10yr	2	50	5	29.4	1	33.3	8	33.3
	>10yr	0	0	11	64.7	2	66.7	13	54.2
Sex	Male	2	50	12	70.6	1	33.3	15	62.5
	Female	2	50	5	29.4	2	66.7	9	37.5
Habitat		0		0.0		0.00	0	0.00	
Urban		3	75	7	41.2	1	33.3	11	45.8
Rural		1	25	10	58.8	2	66.7	13	54.2
Socioeconomic									
	Poor	1	25	9	52.9	2	66.7	12	50.0
	Middle	2	50	5	29.4	1	33.3	8	33.3
	Upper	1	25	3	17.6	0	0.0	4	16.67
	Fistulae/	0	0	2	11.8	1	33.3	3	12.50
Recurrence									
	<6 month	0	0	1	5.9	0	0.0	1	4.17
	6-12 month	1	25	0	0.0	0	0.0	1	4.17
	12-24 month	0	0	1	5.9	0	0.0	1	4.17
	>24 month	0	0	0	0.0	1	33.3	1	4.17
Total	4	100	17	100	3	100	24	100	

**Table-VI**  
*Presentation of the Thyroglossal Duct Cyst according to the age of the children(n=24)*

Character	<5yrs	%	5-10yrs	%	>10 yrs	%	Total	%	
Side	Midline	3	100.00	8	100	12	92.31	23	95.83
	Left	0	0.00	0	0	1	7.69	1	4.17
	Right	0	0.00	0	0	0	0.00	0	0.00
Site									
Suprahyoid		2	66.67	2	25	0	0.00	4	16.67
Juxtra hyoid		1	33.33	5	62.5	11	84.62	17	70.83
Infrahyoid		0	0.00	1	12.5	2	15.38	3	12.50
Sex	Male	3	100.00	7	87.5	5	38.46	15	62.50
	Female	0	0.00	1	12.5	8	61.54	9	37.50
Habitat									
Urban	2	66.67	5	62.5	4	30.77	11	45.83	
Rural	1	33.33	3	37.5	9	69.23	13	54.17	
Socioeconomic							0.00		
	Poor	0	0.00	5	62.5	7	53.85	12	50.00
	Middle	1	33.33	2	25	5	38.46	8	33.33
	Upper	2	66.67	1	12.5	1	7.69	4	16.67
Fistulae/0		0.00	1	12.5	3	23.08	4	16.67	
Recurrence									
	<6 month	1	33.33	0	0	0	0.00	1	4.17
	6-12 month	0	0.00	1	12.5	0	0.00	1	4.17
	12-24 month	0	0.00	0	0	1	7.69	1	4.17
	>24 month	0	0.00	0	0	1	7.69	1	4.17
Total	3	100.00	8	100	13	100.00	24	100.00	

## Discussion

The thyroglossal duct cyst is a well recognized developmental abnormality most often seen as congenital asymptomatic mass of the neck region 7% of the population. While TDCs are most often diagnosed in the pediatric age group, a substantial minority of patients with TDCs are over 20 years of age at the time of diagnosis<sup>22</sup>.

It typically presents as a mobile, painless mass in the anterior midline of the neck, usually in close proximity to the hyoid bone. Less often, TDCs may present with signs and symptoms of secondary infection, or with evidence of a fistula. The standard surgical approach to TDC, encompassing removal of the mid-portion of the hyoid bone in continuity with the TDC and excision of a core of tissue between the hyoid bone and the foramen cecum of tongue, dates back to the late 19th and early 20th centuries and is often referred to as Sistrunk's operation. Malignancy is rarely encountered in TDCs; when such rare tumors do develop (in the order of 1% or so of patients with TDCs), they usually take the form of either papillary carcinoma of thyroid origin, or squamous carcinoma.

The presence of these duct remnants may lead to abnormal phonation and epithelial carcinomas. Therefore, correlation of the rate of thyroglossal duct remnants in a population together with the related clinical symptoms can lead to an early diagnosis and better treatment chances for these problems.

Thyroglossal duct cyst is a developmental disorder of the thyroid where cystic degeneration developed in the persistent thyroglossal duct due to interruption of developing hyoid bone during its descends. It represent the most common congenital anomaly of the neck constitute 70% of all the congenital cervical masses<sup>23</sup> and accounting for 2-4% of all neck masses.

Thyroglossal cysts (TGCs) represent the most common congenital anomaly of the neck, 1 accounting for 2-4% of all neck masses.

### Sex:

It is found equally in both girls and boys.<sup>24</sup>. Males are more commonly affected than female seen in some series<sup>25</sup>. In this series male is more than female (M:F= 1.6:1).

### Age:

In a study conducted by Brousseau V J et al<sup>14</sup> bimodal distribution for age at presentation of

thyroglossal cyst but this finding was not present in our study<sup>26</sup>.

Regarding age of presentation there was a significant difference between the male and female ( $p < 0.05$ ). Female are presented later age (mean  $12.67 \pm 2.40$  years) than male (mean  $7.53 \pm 4.01$  years).

The mean age at presentation in other study other studies were 13.18 years.

On the other hand males were affected more frequently compared to females in our study, ratio of 3.1: 1, which is in agreement with other studies<sup>27</sup>.

### Site:

The lesion usually presents as an asymptomatic painless swelling in the anterior midline or paramidline of the neck<sup>28</sup>, extending from the base of the tongue (foramen cecum) to the sternal manubrium<sup>29,30</sup>. 90% of thyroglossal duct cysts are found in mid line, 10% are to one side of the neck, with 95% percent on the left and 5% percent on the right<sup>31</sup>. Three fourth of the lesions are usually found below the hyoid bone. The cyst may become infected presenting as a painful tense mass with overlying erythematous skin. This may result in sinus formation due to spontaneous rupture or a surgical drainage attempt<sup>32</sup>.

It is frequently presented as midline neck mass that moves with swallowing and protrusion of the tongue.

They may be found anywhere from base of tongue to manubrium<sup>33</sup>. In this study 70.83% cases were in juxtrahyoid (around the hyoid region). In other study Asmatullah et al in pakistan 75.6% were in infrahyoid region<sup>34</sup>. Deaver MJ et al also found most cases inferior to the hyoid bone 65%<sup>35</sup>. Chon SH et al have reported TDC from the mediastinum<sup>36</sup>. Lin ST et al studied patients with left sided TDCs<sup>37</sup> but only one case in this study had left sided neck swelling.

### Presentation

Most of the patient (95.83%) was presented as an asymptomatic midline neck mass which gradually increasing in size. Six (25%) children had globus like sensation and three (12.5%) children had pain due to infection. Only 8.33% children had the complaint of dysphagia or sore throat.

Presenting features may be sore throat, pain, dysphagia, hoarseness, and globus<sup>38</sup>. However, we did not see any case of pain or hoarseness. They may present as infected cyst, discharging sinus, true fistula<sup>39</sup> or intra thyroid cyst<sup>40</sup>.

Less commonly reported manifestations in the literature include acute airway obstruction, misdiagnosed as laryngomalacia in children<sup>41</sup>.

Though the diagnosis is mainly clinical, ultrasonography is of great help. We subjected all cases to ultrasound study and our finding of with homogeneously hypoechoic picture (78.1%) have similar (78.1%) with the study of Asmatullah et al, were different from the study done by Ahuja AT et al, who reported 28% each for anechoic, pseudosolid and heterogeneous picture<sup>42</sup>.

The presence of a solid mass along the thyroglossal duct cyst should raise the suspicion of ectopic thyroid tissue, as it is estimated that 35-70% may contain thyroid tissue in their wall<sup>43</sup>. Rarely, a thyroglossal cyst may contain the only functioning thyroid tissue in the body.

Although only 10% of ectopic thyroid is found in the neck, it may represent the only thyroid tissue in 75% of patients<sup>44</sup>. In this series 4 (16.67%) of the cases were found only thyroid tissue in this remnant.

There is a malignant potential of the dysgenetic thyroid tissue located in a thyroglossal duct cyst; carcinoma develops more frequently in ectopic thyroid tissue than in normal thyroid glands<sup>45</sup>. Carcinoma arising in a thyroglossal duct remnant was first reported in 1925 by Ashurst and White. Most of the patients were in the third through the sixth decades of life. Cancer occurs more frequently in females than in males<sup>46</sup>.

Additional cases of Hürthle cell adenoma and very rarely squamous cell carcinoma were found<sup>47</sup>.

Approximately 85-92% of all thyroglossal duct cyst carcinomas are papillary carcinoma<sup>48</sup>. Although they are characterized by relatively non-aggressive behavior and rare lymphatic spread<sup>49</sup>, the possibility of occult malignancy should be evaluated in such cases.

The presence of calcification in a TGDC on imaging studies suggests papillary carcinoma [because these areas of calcification are psammoma bodies, a histologic hallmark of papillary carcinoma]. About 1% of TDC are found ultimately to harbor thyroid carcinoma which was first reported by Ucherman in 1915.

One of the cases (4.17%) found papillary carcinoma other were reported benign<sup>50</sup>.

Most TGDCs are asymptomatic but could be secondarily infected in an upper respiratory tract infection<sup>51</sup>. Allard (1982) reported that inflammation was the most often mentioned initiating stimulus. Inflammation may lead to a rapid increase in size, cellulitis and even abscess formation<sup>52</sup>. Fistulization onto skin, as seen in two of our patients, occurs in approximately one third of patients<sup>53</sup>. Fistulas may form as a result of spontaneous discharge following inflammation of a cyst, or after surgical intervention, or rarely as congenital fistulas.

### Sonograph

The size of the cyst ranged from 10-30mm in diameter (mean 19.04 SD±4.66).

The typical sonographic description of a TDC has been that of an anechoic, well-circumscribed cyst with increased through-transmission<sup>54</sup>. However, earlier studies in children have shown that most are not simple cysts but instead are either homogeneous or heterogeneous complex hypoechoic lesions<sup>55</sup>.

The pseudosolid appearance of cystic lesions may be due to the proteinaceous content of the fluid, thought to be secreted by the epithelial lining of the cyst. Posterior enhancement suggests the nature of a true cystic<sup>56</sup>.

The pseudosolid appearance of cystic lesions has been described previously for other congenital cystic lesions in the neck, such as branchial cleft cysts and dermoid cysts. The echogenic appearance

is due to the presence of cellular material, cholesterol crystals, and keratin within the cyst<sup>57</sup>.

In this series 33.3% cyst are sonographically anechoic and hypoechoic. One had a hyperechoic solid component. This result is consistent with the study by Ahuja AT et al<sup>58</sup>, where anechogenecity and hypoechogenecity was 28% and 18% respectively. Heteroechogenecity and Psuedosolid was 28%. Posterior enhancement was well defined in 87.50% cases.

All the TDCs in this study were well defined. In 62.5% of the lesions, the walls were thin; 20.83% were thick-walled; and in the remaining 16.6%, the walls were imperceptible. Study of Ahuja AT et al showed 88% have posterior enhancement and 50% had well defined thin walled.

## Conclusion

Thyroglossal duct cyst present as midline neck swelling around the hyoid region. Sometimes it present as globus like presentation and dysphagia. When present with pain it is due to infection.

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