

Thyroglossal Cyst in Children: Presentation, Diagnosis and Management Outcome in the Context of Bangladesh

Syed Hasan Imam Al-Masum¹, Ali Jacob Arsalan²

ABSTRACT

Background and Objectives: Thyroglossal duct cyst (TDC) is considered to be the most common congenital neck swelling in children. But its diagnosis can sometimes be challenging, because of abnormal presentation or associated infection. The aim of the present study is to describe the clinical features, anatomical location and management given to these children in the context of Bangladesh.

Methods: This retrospective review of the past 7 years' (between June 2008 and July 2015) records of all children with a histopathologically confirmed diagnosis of TDC was done at the Department of Pediatric Otolaryngology, Dhaka Shisu Hospital, Sher-e-Bangla Nagar, Dhaka. Thirty patients fulfilled the predefined eligibility criteria and were included in the study. Preoperative evaluation consisted of physical examination with regard to movement of the mass with protrusion of the tongue and ultrasonographic assessment of the mass. All the patients were operated by Sistrunk's approach and outcome was assessed in terms uneventful cure, infection and recurrence.

Result: In the present study the children presented with TDC were on an average 7.1 (range: 3.5 - 11 years) years old and were predominantly girls (60%). Majority of patients belonged to middleclass (70%). Urban and rural representations were almost equal. Half of the patients were diagnosed between 5 - 10 years of age. Majority (80%) of the TDCs were located in infrahyoid region just lateral to the midline, 16.7% were located close to the hyoid bone (juxtahyoid) and 3.3% to the suprahyoid region in the midline. All the 30 cases presented with painless cystic swelling which moved upwards with protrusion of tongue. No signs of infection or associated diseases (myxoedema or thyroglossal fistula) were present. An anechoic character with absence of fluid level was detected with ultrasono imaging of the mass.

Conclusion: Clinical presentation and anatomical location and management were almost same as elsewhere in the world. But in the absence of a complete database including the follow up data, it is not feasible to plan for further improvement of the children with TDC.

Key words: Thyroglossal cyst, Sistrunk operation, children, presentation, diagnosis, management, outcome.

INTRODUCTION

Thyroglossal duct cyst (TDC) is one of the most common congenital neck masses occurring in children and represents approximately 70% of the congenital anomalies that occur in the neck.^{1,2}

The TDC results when thyroglossal duct persists and fails to obliterate along any portion of the thyroid's descent to the pretracheal position in

the midline of the neck.^{2,3} In most circumstances, a clinical history and physical examination are sufficient to make a correct preoperative diagnosis as majority of patients present commonly with an asymptomatic cystic neck mass in the midline near the hyoid bone (Fig 1 & 2).⁴ However, in some cases, the diagnosis of TDC is not clinically apparent and requires the help of preoperative imaging. And elective surgical

Authors' Information:

¹ Dr. Syed Hasan Imam Al-Masum, Associate Professor, ENT Department, Dhaka Shisu Hospital, Sher-e-Bangla Nagar, Dhaka.

² Dr. Ali Jacob Arsalan, Resident Medical Officer, ENT Department, Dhaka Shisu Hospital, Sher-e-Bangla Nagar, Dhaka.

Address of Correspondance: Dr. Syed Hasan Imam Al-Masum, Associate Professor, ENT Department, Dhaka Shisu Hospital, Sher-e-Bangla Nagar, Dhaka, **Cell:** +8801711-141543, **E-mail:** shmasum_ent@yahoo.com



FIGURE 1: Thyroglossal cyst presenting as swelling in the neck near midline (Adapted from Mandhanet al., 2015)⁹

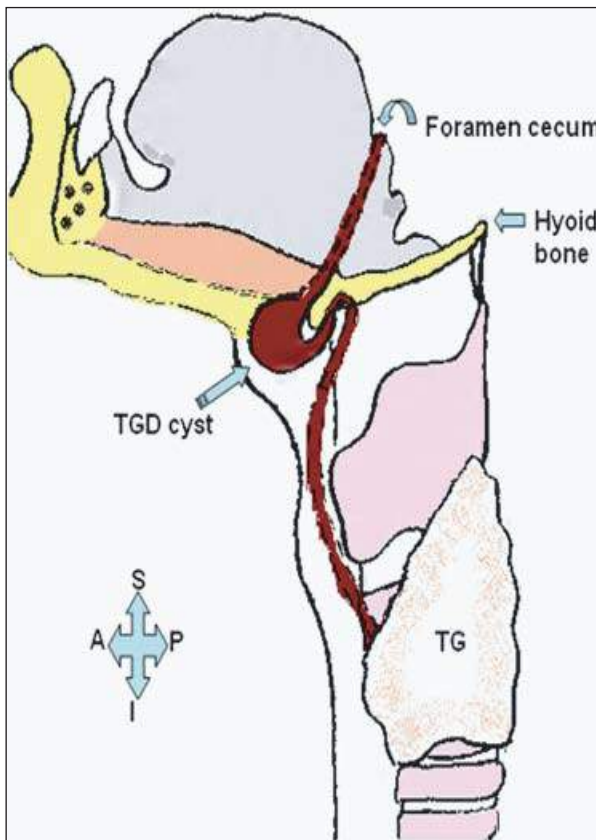


FIGURE 2: Schematic representation of the thyroglossal duct (TGD) extending from the region of foramen caecum (curved arrow) upto the thyroid gland (TG). Note the close relationship of TGD cyst (broken arrow) with the hyoid bone (solid arrow) (Adapted from Gaddikeri et al., 2014)¹⁰

excision (Sistrunk procedure Fig.3) is the treatment of choice for uncomplicated TDC to avoid the risk of infection. When thyroglossal duct cysts (TDCs) present with an infection, treatment includes antibiotic, aspiration/drainage followed by surgery. Successful treatment of TDC is often measured by the recurrence rate. Recurrence of TDCs after the Sistrunk procedure is reported to be from 2.6 - 5%.^{3,5-8}



FIGURE 3: Removal of the cyst by Sistrunk's approach (Adapted from Mandhan et al., 2015)⁹

Other attributed factors for recurrence include pre-operative infection, inappropriate initial surgery, surgeon's skill (level) and abnormal presentation/anatomy.^{1,11-14} We describe our experience for the management of TDCs over a 7-year period. We have also specifically looked for complications, recurrence rate and possible attributing factors for recurrent TGDCs in children in Dhaka Shishu Hospital.

MATERIALS & METHODS

Having obtained approval from the Institutional Review Board of Dhaka Shisu Hospital, we performed a retrospective review of the past 7 years' (from June 2008 to July 2015) records of indoor patients with a diagnosis of TDC at the

Department of Pediatric Otolaryngology. The inclusion criteria were age 12 years or younger and patients in whom TDC was diagnosed clinically and was confirmed histopathologically after surgical excision. The histopathological criteria for diagnosing TDCs were presence of ductular or cystic structures lined with cuboidal or columnar epithelium or non-keratinizing stratified epithelium without epidermal appendages. However, patients who had a recurrent TDC were excluded. Thirty patients fulfilled the eligibility criteria and were included in the study. Age, sex, anatomical location, clinical presentation, age at diagnosis, associated diseases (myxoedema or thyroglossal fistula) and ultrasonologic finding were noted. Preoperative evaluation consisted of physical examination with regard to movement of the mass with protrusion of the tongue and ultrasonographic assessment of the mass. All the patients were operated by Sistrunk's approach with dissection of the cyst with excision of part of the hyoid bone centrally and coring out muscle attached to the hyoid bone to a level near foramen cecum. In no case the mouth was entered.

RESULTS

Age distribution of the children shows that 50% of the patients were diagnosed between 5 - 10 years of age, 30% below 5 years and 20% at 10 or > 10 years of age. The mean age of the children was 7.1 (range: 3.5 - 11) years. The children were predominantly girls (60%). In terms of socioeconomic status of the patients lower middle class (30%) and middle class together comprised more than two-thirds (70%) of the patients. The poor and the upper middle class formed 20% and 10% of the cases respectively. Urban and rural residents each were 40% (Table I). Anatomical location shows that majority 24(80%) of the TDCs were located in infrahyoid region just lateral to the midline (paramedian location), 5(16.7%) were located close to the hyoid bone (juxtahyoid) and 1(3.3%)

to the suprahyoid region in the midline (Fig. 4). All the 30 masses were cystic in feeling with no complain of pain. The mean size of the cyst was

TABLE I. Distribution of patients by their demographic characteristics (n=30)

Demographic characteristics	Frequency	Percentage
Age (years)		
<5	9	30.0
5 - 10	15	50.0
≥ 10	6	20.0
Sex		
Male	12	40.0
Female	18	60.0
Socio-economic status		
Poor	6	20.0
Lower middle	9	30.0
Middle	12	40.0
Upper middle	3	10.0
Residence		
Urban	12	40.0
Rural	12	40.0
Slum-dweller	6	20.0

*Mean age = (7.1 ± 2.5) years; range = (3.5 -11) years

ORIGINAL ARTICLE

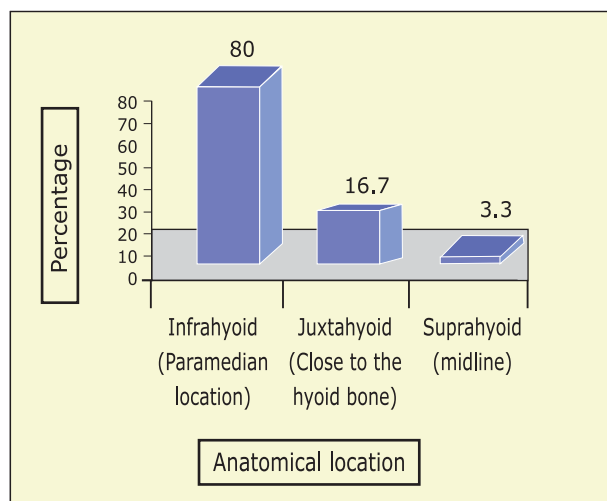


FIGURE 4: Distribution of patients by anatomical location

1.6 cm. The swelling moves upwards with protrusion of tongue. No signs of associated infection were evident on clinical examination (Table II). Associated diseases like myxoedema or thyroglossal fistula were also not found. An anechoic character with absence of fluid level was evident with ultrasonogram imaging of the mass (Table III). Histopathological evaluation revealed findings consistent with the presence of thyroglossal remnant in all cases. The cystic and/or ductular epithelium was ciliated in 8, nonkeratinizing in 12 and mixed in 10 specimens. Multiple ductular structures were found in 26 specimens (85%). The lengths of resected hyoid bones ranged from 7 mm to 19 mm (average 12.5 mm). No ectopic thyroidal tissue was seen.

TABLE II: Distribution of patients by clinical presentation (n=30)

Clinical presentation	Frequency	Percentage	Mean \pm SD
Cystic mass	30	100.0	---
Pain	00	0.0	---
Size (cm)	---	---	1.6 \pm 0.2
Moves upwards with protrusion of tongue	30	100.0	---
Associated infection	00	0.0	---

TABLE III: Distribution of patients by image findings (USG) of (n=30)

Image findings (USG)	Frequency	Percentage
Anechoic	30	100.0
Fluid level seen	00	0.0

DISCUSSION

The present study demonstrated that the mean age of the children presented with TDC was 7.3 (range: 4 - 11) years with 40% of the children ranging from 5 - 10 years. This finding is almost consistent with the data available from the Arabian Peninsula, where mean age at the time of surgery was 7.6 years with half of the patients

being diagnosed between 5 - 10 years of age.⁹ While in our study the children were predominantly girls (60%), the later study showed a male preponderance.⁹ A study conducted in Korea on 16 children also have the similar age distribution [mean age 6.4 (2 - 10 years)] but males were somehow more (56.2%) than the females (43.8%).¹⁵ In a Turkish study, Turkyilmaz and associates demonstrated mean age of the 27 patients to be 6.25 years (range: 1.5-14 years) with boys being higher (55.5%) than their girl counterparts (45.5%).¹⁶

Anatomical location shows that majority (80%) of the TDCs were located in infrahyoid region just lateral to the midline (paramedian location), 16.7% were located close to the hyoid bone (juxtahyoid) and 3.3% to the suprahyoid region in the midline. Consistent with these findings Lee showed that 8(50%) were located in the midline and 8(50%) just to one side of the midline. Twelve (75%) were infrahyoid in location, 4(25%) were at the level of the hyoid, and none was suprahyoid.¹⁵ However, in contrast to these findings Mandhan's study⁹ demonstrated that all TDCs were situated at midline. In Turkyilmaz's study¹⁶ the majority (81%) lesions were localized in the midline at the hyoid level. Most TDC cysts are located either at or below the level of the hyoid bone, with 50% at hyoid bone and 20-25% in suprahyoid neck, often in the midline. The remaining 25% are located in the infrahyoid neck where they are in the midline or within the strap muscles in a para midline location.¹⁷ However, Athow and colleagues described that though majority of TDC lies close to the hyoid bone, they can be at any site along the pathway of descent of the thyroid anlage.¹⁸

All the cases in our study presented with painless cystic swelling which moves upwards with protrusion of tongue. No signs of infection or associated thyroid diseases were evident. Studies show that TDCs most often present with a painless swelling, a draining sinus or a tender mass.¹⁹

Movement of the cyst with swallowing is often cited as a reliable diagnostic sign but can be difficult to evaluate in young children and misdiagnosis could occur due to the similar manifestation of the dermoid cyst, lymphadenopathy and cystic hygroma when located near the hyoid bone.¹⁹⁻²² Unusual locations of TDCs have also been described.²³⁻²⁵ Embryologic development of neck structures is a reason that these remnants can be found in other locations.²⁶

In all patients, an anechoic character with absence of fluid level was detected with ultrasonogram imaging of the mass. In Turkyilmaz's study¹⁶ ultrasonographic examinations were consistent with anechoic pattern in 7(31.8%), pseudosolid in 9(40.9%) and heterogenous in 6(27.2%) cases. In Madhan's study 71 children had an ultrasound done before surgery and it was sensitive in 95% cases. Our results are almost similar to the results of the 3 other studies.²⁷⁻²⁹ Comparing sonographic evaluation of TDCs in children. Preoperative imaging is important to confirm the diagnosis, to identify the presence of functioning thyroid tissue in the neck, and to detect any possibility of malignant change within the cyst.³⁰

The procedure popularized by Sistrunk has been the most commonly used in the treatment of TDC, with very low recurrence rates (3%).^{20,31} In our study no recurrence was reported indicating that recurrence was either absent or the recurrence cases took treatment from elsewhere. The factors held responsible for recurrences comprised variable entities such as dermal involvement, patients age (childhood), rupture of the cyst during the operation, lobulation of the cyst, inflammation and/or infection, elimination of the coring-out procedure in an attempt to follow the suprahyoidal tract, and cases with fistulas.^{32,33} Ein et al.³⁴ had considered infection and fistulas as being responsible for recurrence in

their large series of 270 cases. Solomon and Rangecroft, in contrast, had suggested inappropriate surgical approach due to initial misdiagnosis rather than infection and/or inflammation as the reason for recurrence. We agree with LaRiviere et al⁴ that formal incision and drainage should be avoided, if possible, to prevent seeding of ductal cells outside the cyst, which increases the risk of recurrence. As follow up data of the children who underwent operation in our hospital are not available, it is not feasible to comment on the postoperative infection rate of these patients in our setting.

CONCLUSION

This is, by far, the first reported series of TDCs in children from Dhaka Shisu Hospital, Bangladesh. Clinical presentation and anatomical location and management are almost same as elsewhere in the world. But in the absence of a complete database including the follow up data, it is not feasible to plan for further improvement in management of the children with TDC. We suggest keeping a complete digital database including the follow up data so that the data can be extrapolated to planning and decision making in the better management of TDC cases.

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