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ORIGINAL ARTICLE

A Decade of Experience of Management of Thyroglossal Duct Cyst in a Tertiary Care Hospital: Differentiation Between Children and Adults

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Abstract Variations in thyroglossal duct cysts (TGDCs) between children and adult are mentioned very little in literature. The lesion mostly found in children but adult population also possesses this anomaly. The aim of this study was to determine the differences in clinical presentations and surgical outcomes of TGDC between children and adults. A retrospective chart review of all patients with TGDCs managed in our hospital from July 2004 to June 2014. All records were reviewed for age, sex, location of cyst in neck and with relation to hyoid bone, size, postoperative complication and recurrence rates. Differences between children and adults were assessed. A total of 39 patients (21 children and 18 adults) were treated for TGDC. Of the pediatric group, 71.4% were male and 28.5% were female, whereas 72.2% of the adults were male and 27.7% were female. Adults were more likely to develop other complaints like neck pain, dysphagia and dyspnea. Position was almost similar in both age groups with midline and infrahyoid location while laterality was seen in adult only. Size of Cyst was found to be larger in adults. The recurrence and post operative complication rates between children and adults were not significantly different. TGDC has male predominance. Clinical presentations were almost similar in both age groups. Although lateral deviation, increase size of cyst and recurrences were seen in adults only, Sistrunk procedure is recommended as a safe and standard surgical treatment in both age groups.

Keywords Thyroglossal duct cyst · Difference · Clinical presentation · Surgical outcome · Children · Adult

Introduction

Thyroglossal duct cysts (TGDCs) represent the most common midline congenital neck masses. The cyst accounts for approximately 70% of all congenital neck abnormalities [1]. Around 60% of the lesions are found before age 20 [2], in contrast to 7% [3] of the adult population who possess this anomaly. In one of the largest series by Hsieh et al. [4] regarding pediatric congenital cervical cysts, it was found that this abnormality mostly occurred in males. The cyst presents as a firm, well circumscribed, nontender mass moving upward with protrusion of tongue and swallowing, as it attaches with foramen caecum through embryonic duct. Most thyroglossal duct cysts are positioned in the midline [4, 5] and they can be located anywhere between foremen caecum to the suprasternal region but most of them found below hyoid bone "infrahyoid type" [6].

Schlange in 1893 described the treatment of TGDCs by removing the central portion of the hyoid bone along with the cyst, which results in higher recurrence rate [7]. In 1920, Walter Ellis Sistrunk proposed the surgical management of this congenital abnormality by doing an en bloc cystectomy, central hyoidectomy, and tract excision up to the foramen caecum, which reduces recurrence rate significantly [8, 9] and this remains the standard surgical approach till today. Although large amount of literature

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encouraging the Sistrunk approach as a surgical management, there are reports of postoperative complication and recurrence in both children and adults from this treatment.

The differences in clinical presentation and surgical outcomes of thyroglossal duct cyst between children and adults are mentioned very little in literature. Hence we performed a retrospective study to determine the differences in clinical presentation, sex, location of cyst, post operative complication and recurrence rates of TGDCs in children and adults. These results will be beneficial in predicting outcome and optimizing management of TGDCs in both.

Patients and Methods

A retrospective chart review was performed to evaluate patients with a diagnosis of TGDCs in the Department of Otorhinolaryngology and Head–Neck surgery, Aga Khan University Hospital, from July 2004 through June 2014.

A total of 50 patients with preoperative diagnosis of TGDCs were identified in the medical records within this Period. After reviewing surgical pathology reports, we excluded 11 patients with misdiagnoses. The remaining 39 patients were recruited and divided into 2 groups: children (<15 years) and adults (≥15 years). Age, sex, location of cyst in neck and with relation to hyoid bone, size, postoperative complication and recurrence rates were reviewed from chart.

SPSS version 21 was used for data management and statistical analysis. Continuous variables were presented as mean ± SD. The significance of the difference in clinical characteristics between children and adults was calculated by χ^2 test (or Fisher exact test) for categorical variables and Student *t* test for continuous numerical variables. A *p* value of <0.05 was considered statistically significant. As this was a retrospective study of routinely collected clinical data, formal consent was not required.

Results

Age and Sex

Of the 39 patients, 18 were adults and 21 were children; age ranged from 1 to 50 years. In the adult group, there were 13 males and 5 females, and in the pediatric population, there were 15 males and 6 females. There were more males (71.7%) as compare to females (28.2%) in the overall population. The mean age of the children was 8.2 ± 3.8 years and the mean age of the adults was 29.6 ± 12.4 years. The average age at diagnosis was 18.1 ± 13.9 years in the overall population.

Table 1 Presenting symptoms of TGDCs

	Children (n = 21)	Adult (n = 18)
Neck mass	21 (100%)	18 (100%)
Neck discharge	7 (33.3%)	7 (38.8%)
Neck pain	0	3 (16.6%)
Dysphagia	0	3 (16.6%)
Dyspnea	0	2 (11.1%)

Table 2 Location of cyst in neck and in relation to hyoid bone

	Children (n = 21)	Adult (n = 18)
Location in neck		
Middle	21 (100%)	14 (77.7%)
Left	0	2 (11.1%)
Right	0	2 (11.1%)
Location in relation to hyoid bone		
Infrahyoid	10 (47.6%)	8 (44.4%)
Suprahyoid	3 (14.2%)	5 (27.7%)
Overhyoid	8 (38%)	5 (27.7%)

Presenting Symptoms of Thyroglossal Duct Cysts

The most common clinical presentation of TGDC is neck mass which is present in all of our cases in both children and adults. In both groups, 7 (33.3%) children and 7 (38.8%) adult patients presented with neck discharge. Only adult as compare to children, were present with a complaint other than mass or discharge, including neck pain, dysphagia and dyspnea, which accounts for <20% as shown in Table 1.

Location in Neck

The most common location in neck in both groups were midline but in adults they were also found laterally (22.2%) to either right or left (Table 2). When we compare this laterality with age, the *p* value is found to be statistically significant ($p = 0.037$).

Location in Relation to Hyoid Bone

Infrahyoid TGDC occurred more frequently in both groups (46.1%) followed by overhyoid (33.3%) and suprahyoid (20.5%) in our cohort as shown in Table 2.

Preoperative Evaluation

The preoperative assessment included physical examination, serum thyroid function tests (TFTs), ultrasonography of the neck, computed tomography (CT), magnetic

Table 3 Preoperative studies in different age groups

	Children (n = 21)	Adult (n = 18)
Physical examination	11 (52.3%)	11 (61.1%)
Ultrasound	8 (38%)	3 (16.6%)
TFTs	2 (9.5%)	1 (5.5%)
CT	0	1 (5.5%)
MRI	0	1 (5.5%)
FNAC	0	1 (5.5%)

Table 4 Size of cyst in different age groups

	Children (n = 21)	Adult (n = 18)
Size of cyst (cm)		
Mean	2.1 ± 0.9	4.6 ± 1.2
Range	1.7–5	2–7

resonance imaging (MRI), and fine-needle aspiration cytology (FNAC). The most frequent preoperative study was physical examination, which was more than 50% in both groups (Table 3). Ultrasonography was the second most common modality in both groups (28.2%) followed by thyroid function tests (7.6%). Neck CT scan, MRI and FNAC examinations were only performed in the adult group.

Surgical Management

All of our patients underwent Sistrunk's procedure as a primary modality of treatment. All operations were performed by experienced surgeons (assistant professor or above) and surgical specimens were sent for histopathologic examination. A penrose drain was placed and all patients received postoperative antibiotic therapy. Follow-up ranged from 6 to 12 months (mean = 8.6 ± 2.0 months).

Size of Cyst in Final Histopathology

In children, mean of size of cyst was 2.1 ± 0.9 cm (1.7–5 cm), whereas in adults it was 4.6 ± 1.2 cm (2–7 cm). There was statistically significant difference ($p = 0.001$) on comparing mean of size of the cyst with that of mean of age showing that greater the age, larger will be the size of the cyst (Table 4).

Postoperative Complication and Recurrence

The preoperative diagnosis of TGDC was histopathologically established postoperatively in all 39 cases. Complications associated with surgery found to be rare. There were no major complications resulting from Sistrunk procedure. Local wound infection found to be most common minor complication in both groups affecting five (12.8%) children and two (11.1%) adults. In children, apart from wound infection, two (9.5%) cases were also had seroma formation. There were only two (5.1%) patients of adult group in our series in which recurrence was noted and it was recognized within 6 months of surgery. Both presented with an infected neck mass and were treated with a repeated Sistrunk procedure or extended Sistrunk procedure. Although the recurrence rates were not statistically different between children and adults ($p = 0.20$) but only the adult population had recurrence of the disease.

Discussion

Thyroglossal duct cysts (TGDCs) are the most frequent midline congenital neck cyst. In two of the largest series regarding congenital neck masses, TGDC accounts for more than 50% in both studies [4, 10]. They can be detected at any age but since it is a congenital abnormality, it is anticipated that the cyst preponderate in children. Meta-analysis by Allard [11] showed higher incidence of TGDC in children as compare to series by Brousseau et al. [12] in which adults have more this malformation. Our series shows a slightly higher manifestation in children (53.8 vs. 46.1%). The dissimilarities in presentation may be due to whether the age was recorded at the time of onset of symptoms or at the time of initial diagnosis.

Dispute about the gender distribution of TGDCs also exists in the literature. Most of the studies reported an equal distribution of TGDCs among males and females [6, 11, 12]. While Hsieh et al. [4] and Turkyilmaz et al. [7] observed male and female predominance respectively. In our series, we observed male predominance in both groups. These contrasts may be due to genetic and geographic differences.

Thyroglossal duct cysts mainly presents as a mass or infection. In case of mass, it is solitary, midline and moving with swallowing and if infected, patient presents with discharging neck fistula, neck pain, dysphagia and dyspnea. In our study, all patients presented with neck mass in both groups. Apart from mass and neck discharge, other symptoms only manifest in adults showing similar result as Brousseau et al. [12].

Majority of TGDCs are situated in midline of neck while 10–24% are located laterally, mostly towards left [3, 5]. It is obvious that lateral presentation creates other differential diagnosis of neck in mind e.g. branchial cyst, laryngocele, cystic hygroma etc. In this series, midline demonstrates the common location in neck in both groups. All children in our series having midline presentation while adults are the

only class in which lateral deviation of mass found, similar with Shih-Tsang study [6].

In Allard series [11] and Brousseau et al. [12], infrahyoid position is the most common location of TGDC in relation to hyoid bone. There was adult predominance of infrahyoid location as compare to children in Lin et al. [6] and Ahuja et al. [13] reviews. Similarly in our series, infrahyoid TGDC occurred more frequently overall and also dominate in both groups.

Association of lesion with hyoid bone along with midline presentation and movement with deglutition is often cited as a reliable diagnostic sign. Good clinical history and head and neck examination are adequate to make a correct preoperative diagnosis. In our series, most of the cases diagnosed with it, similar with Davenport review [14]. However in some cases with unusual presentation of TGDC, preoperative diagnosis becomes difficult. In our series, apart from clinical diagnosis, five other diagnostic modalities were used in the preoperative evaluation i.e. ultrasonography, CT scan, MRI, FNAC and serum thyroid function tests. Pre-operative imaging such as ultrasound, neck CT and MRI used to find the existence of normal thyroid tissue while FNAC performed to rule out thyroglossal duct carcinoma in cyst which is <1% of cases [15, 16].

Ultrasound was the second common investigation in both groups because it is noninvasive, inexpensive, and does not involve ionizing radiation or sedation, which is particularly important in children. CT and MRI play an accessory role to more accurately outline the anatomy of large cysts and require sedation was only performed in adults in our study. There was only one case of adult group in which FNAC was performed because there was positive family history of thyroglossal duct carcinoma.

Before 1893, a simple incision and drainage was the treatment of choice for TGDCs, resulting in inappropriately high recurrence [17]. Schlange then proposed the method of removing mid portion of hyoid bone along with cyst, a procedure that had a recurrence rate of 20% [7]. After the Sistrunk operation introduced in 1920, the recurrence rate reduces to 3–6% [6, 12] and it is the standard surgical technique performed for TGDC even today. In our series, only 2 adult patients had recurrence. Likely possibility of recurrence in adult may be larger cyst with inadequate removal tract. Both cases treated with extended Sistrunk procedure with no recurrence. The recurrence rate in children and adults are not significant in this review similar with the literature [6, 12].

Size of cyst varies according to age. In our study, the cyst was found to be larger in adults than in children, which may point toward that the lesion increases with advancing age.

Sistrunk operation is relatively a safe procedure with lesser complication rates. There are chances of both major (abscess, hematoma, nerve palsies, hypothyroidism) and minor (local wound infection, seroma, wound dehiscence) complications. Major complications are extremely rare as compare to minor ones. Some reviews [6, 18] reported minor complications found frequently in adults in contrast to other [12, 19] in which younger group suffered more. Our series showed consistent results with latter reviews that pediatric population having more complications but this difference was not significant.

Conclusion

Our results indicate that thyroglossal duct cyst has male predominance. Apart from mass and neck discharge, adults were more likely to develop other complaints like neck pain, dysphagia and dyspnea. Position was almost similar in both age groups with midline and infrahyoid location while laterality was not seen in children. The lesion was larger with the advancing age. Meticulous history and examination is sufficient for preoperative diagnosis while imaging modalities are useful to rule out other differentials of neck. Sistrunk procedure is recommended as a safe and standard surgical treatment as the recurrence rates and complications between children and adults were not significantly different.

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Compliance with Ethical Standards

Conflict of interest None of the authors of this paper has a financial or personal relationship with other people or organizations that could inappropriately influence or bias the content of the paper.

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