Original paper

Pathological Analysis of Thyroglossal Duct Cyst in Children

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Abstract

Background: Thyroglossal duct cysts (TGDCs) are the most common form of congenital neck cyst, accounting for up to 70% of such lesions. There has been no consensus on which factors predict outcome of thyroglossal duct cyst excision.
Objective: To evaluate the relevance of symptomatology, age at presentation and the histopathology of TGDCs.
Methods: Retrospective review of patients with TGDC at Al-Hissain medical city. Data collected included patient's age, gender, clinical presentation, presence or absence of preoperative infection, size and location of the lesion,
Results: Thirty –one patients were identified (71% female and 29% male). Age of presentation was bimodal and ranged from few months to 17 years. The most common presenting symptom was the presence of an asymptomatic midline neck mass (71%). A history of preoperative TGDC infection was present in 3% of patients ≤2 years of age and in 10% of patients ≥5 years of age.
Conclusions: The finding of a midline neck mass is the most common presentation of TGDCs in toddlers, whereas infection is the most common presenting symptoms in school-aged children. Complete and precise clinical information is a prerequisite in order for pathologists to make accurate diagnoses of congenital cervical cysts.
Keywords: *Thyroglossal duct cyst; Neck mass; histopathology; Symptoms.*

Introduction

Thyroglossal duct cysts (TGDCs) are the most common common cervical cyst, as it occurs in about 7% of the population [(1)] and constitutes 70% of the congenital malformations that affect the cervical area [(2)].

They commonly present in children or adolescents, yet up to one-third of patients are 20 years or older. Males and females are equally affected [(3)]. In most cases, a meticulous clinical history and physical exam are sufficient to make the diagnosis.
The true incidence is unknown, and, in fact, many are never detected clinically. Ellis and van Nostrand found a 7% incidence of TGDCs in postmortem study of 200 adults [(4)]. Despite the relative frequency of TGDCs, familial occurrences are rare. The inheritance pattern is usually autosomal dominant with incomplete penetrance, and a female preponderance has been noted [(5)].

Embryology

The thyroid analage develops from an epithelial proliferation that originates at the junction of the anterior two-thirds and posterior-third of the tongue, the future site of the foramen cecum. At 4 weeks gestation, a ventral diverticulum forms and descends in the midline of the neck as the thyroglossal tract [(6,7)]. Maintaining its attachment to the base of the tongue, the tract travels anterior to the hyoid bone and then in front of the thyroid cartilage, cricothyroid membrane, and cricoid cartilage [(4)].
The thyroid assumes its final position anterior to the tracheal rings, where the lobes separate by 7 weeks gestation. Normally, the tract atrophies and disappears by 10 weeks gestation

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[6,7].Portions of the tract and thyroid tissue remnants may persist at any point along the migratory path. Occasionally, there is no migration, and the entire thyroid is arrested at the base of the tongue resulting in a lingual thyroid [8]. TGDC result from expansion of a remnant of the thyroglossal tract. The cause of expansion is unknown; however, inflammation is the most likely etiology. Adjacent lymphoid tissue reacting to infections may stimulate the epithelial remnants to undergo cystic changes [3].

Clinical presentation

Patients with a TGDC typically present with a midline upper neck mass that moves with tongue protrusion and swallowing. The cyst is generally nontender and may reach a diameter of up to 10 cm. While most lies in the midline (fig.1), they may less commonly be located laterally, usually to the left [3]. Patients may report a recent upper respiratory tract infection, which can cause a sudden increase in the size of the mass [10].

The position of the TGDC can be described as lying in one of the following four locations: (1) intralingual, (2) suprahyoid, including submental, (3) thyrohyoid, and (4) suprasternal. In approximately 60% of cases, the cyst is found intimately associated with the hyoid bone adjacent to the thyrohyoid membrane [3].

Although most TGDCs are asymptomatic, those found intralingually may cause choking sensations, dysphagia, and dysphonia [3,11]. A cutaneous or oral fistula is a secondary feature of the disease and may result from suppuration, trauma or inadequate surgery of a thyroglossal cyst [3,7,11].

Adults are more likely than children to present with a complaint other than mass or infection, including pain, sore throat, dysphagia, hoarseness, and fistula formation [12]. It is unclear if adolescents present differently than infants

The differential diagnosis of a TGDC includes dermoid cysts, sebaceous cysts, branchial cleft cysts, thyroid nodules, lymphadenopathy, and lipomas [10,11]. In 1—2% of patients with suspected TGDCs, the mass is actually an ectopic thyroid gland [13]. Preoperative detection of hypothyroidism or absence of thyroid tissue in the normal site may aid in making this diagnosis [13,14]. Thyroglossal duct carcinomas are rare and occur in approximately 1% of TGDCs. They are usually not predicted upon presentation and are incidentally diagnosed after surgery. The vast majority has a papillary thyroid origin, but there are rare reported cases of squamous cell carcinoma [15].

Diagnosis

The epithelial lining of a TGDC ranges from squamous epithelium to pseudostratified ciliated columnar epithelium. Salivary gland tissue or thyroid gland tissue may be seen in the wall of the cyst.

The cyst fluid can be described as mucoid, gelatinous, or purulent yellowish-white to dark brown that may contain cholesterol[3].

Preoperative imaging is important to confirm diagnosis, identify normal functioning thyroid tissue, and to detect malignancy. High-resolution ultrasound remains the ideal initial imaging modality for TGDCs. It is easily accessible, inexpensive, non-radiating, and does not require sedation [14,16]. An uncomplicated cyst will appear anechoic and well circumscribed or pseudosolid from proteinacious fluid contents. Doppler may identify any solid component suggestive of malignant disease.

Routine radioisotope thyroid scanning is not necessary if a normal thyroid gland is demonstrated on preoperative ultrasound since the cyst is unlikely to contain the only functioning thyroid tissue [14,16].

Other modalities that have been employed for the diagnosis of TGDCs include computed tomography (CT) or magnetic resonance imaging (MRI).

Management

There is no effective medical therapy for the treatment of TGDCs; the use of

sclerosing solutions and coagulation are not recommended unless surgery is refused [3]. The standard surgical procedure, described by Sistrunk in 1920, aims for removal of all possible thyroglossal tract remnants to prevent recurrence. In this procedure, the midline portion of the hyoid bone is resected along with a wide core of tissue belonging to the midline area between the hyoid and foramen cecum [17]. The Sistrunk procedure is associated with a very low morbidity. In a retrospective review of 35 patients, Maddalozzo et al. reported no major complications following surgery and only minor wound-related complications in 29% of patients [16].

Of most concern after resection is recurrence of disease due to incomplete removal of tract remnants. Using Sistrunk's technique on 270 patients, Brown and Judd were able to reduce the recurrence rate to 4% [18]. Simple cyst excision is associated with a recurrence rate of up to 40% [11]. The best chance of curative resection is at initial presentation [7]. Although many have reported preoperative cyst infection as a significant risk factor for recurrence, [19]. there has been no consensus on which factors predict outcome of thyroglossal duct cyst excision.

Materials and methods

A retrospective analysis of the computerized Systematized files of all specimens accessed in the department of Pathology at Al-Hussain medical city were searched from the inception of the file in January 1, 2012 to December 30, 2016. Thirty -one patients met inclusion criteria for this study and had a postoperative diagnosis of ''thyroglossal duct cyst''. The study population was confined to the pediatric age group under the age of 18 years. There were 31 patients including 22 girls and 9 boys Age at diagnosis ranged from neonate to 18 years. The histology of all cases was reviewed and correlated it with the clinical information in the medical records.

Results

Of the 31 patients, 22 (71%) were female and 9 (29%) were male. Table (1). Four patients (13%) were defined as infants (<2 years), while 21 (68%) patients were defined as school aged (>=5 years) at presentation with the remaining patients falling in between as shown in table. 2.

Presenting symptoms are displayed in Table (3), however, 71% presented with Midline neck mass,16% presented with thyroglossal duct cysts Moves with tongue protrusion and/or swallowing as in and 13% presented with preoperative infection .

Of the four patients in the infant age range, one (3%) had a positive history of preoperative infection while three of the 14 school-aged children (10%) had a positive history of preoperative infection (Table 2). Imaging studies were obtained in all cases

The microscopic examination revealed a cystic structure with lumen lined by newly formed connective-vascular tissue with edema and numerous inflammatory polymorphic elements in addition to hemorrhagic microfoci. The connective wall has rare inflammatory elements. Bundles of striated muscle fibers were detected on the periphery. Pieces of hyaline cartilage and bone made up of bone lamellae and hematogenic bone marrow surrounded by tube-like structures lined by pseudo-stratified epithelium and by stratified squamous epithelium (fig. 2,3,4); an area with bone lamellae hosting a polymorphonuclear neutrophil (PMN) exudates was also noted.

They were all located on the midline at or immediately adjacent to the hyoid bone. Three of them were initially diagnosed as benign cysts.

However, their microscopic features showed cystic lesions lined with squamous

or denuded epithelium with fibrosis, and both acute and chronic inflammation.
We reclassified them as thyroglossal duct cysts due to their typical location at the hyoid bone as recorded on the medical charts. As shown in table (4).
Complete excision of the cyst is advocated to avoid the complications of infection, which will make resection more difficult and recurrence more likely [(2)]. Thus, it is important for the pediatrician, pediatric surgeon, and pathologist to be familiar with the embryologic origin and differentiation of cystic lesions in order to accurately diagnose and guide further therapy.

Discussion

Thyroglossal duct cysts are the most common malformation found in the neck which constitute one of the most intriguing areas of pediatric pathology. The differential diagnosis is based upon the location and histology of the epithelium and the surrounding stroma.

Table 1. Gender ratio of the cyst

Type of cyst	No. of cases	Gender ratio	
		Female (%)	Male (%)
Thyroglossal duct	31	22(71%)	9(29%)

Table 2. Preoperative infection distributed by age

Age at operation	(no. of patients)	(%)	Number with preoperative infection	(%)
≤ 2	4	13	1	3%
3-4	6		0	0
≥ 5	21		3	10%
Total	31	100	4	13%

Table 3. Clinical presentation of thyroglossal duct cysts

Clinical presentation of thyroglossal duct cysts	
Symptom	Number (%)
Midline neck mass (no infection or drainage)	22 (71)
Moves with tongue protrusion and/or swallowing	5(16)
Infection	4(13)

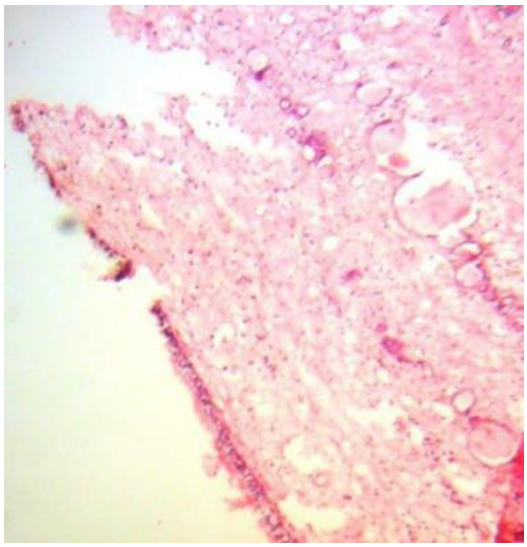

Figure 2. Cyst showing pseudo-stratified columnar epitheliumand thyroid follicles in the wall; H & E x 100 magnification.

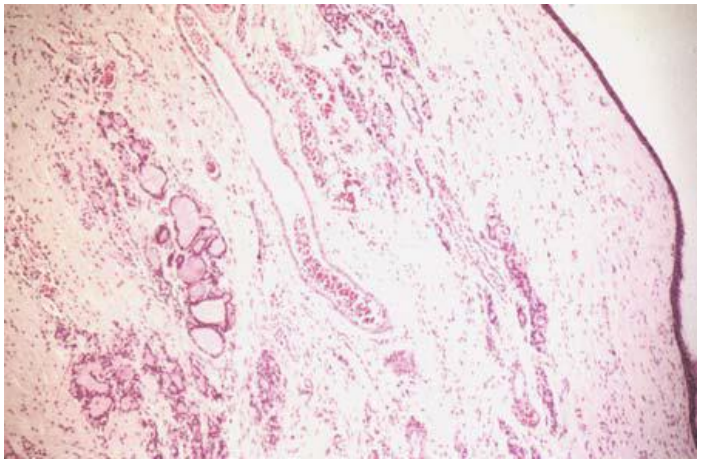

Figure 3. Thyroglossal duct cyst. Th yroid follicles are present in the wall

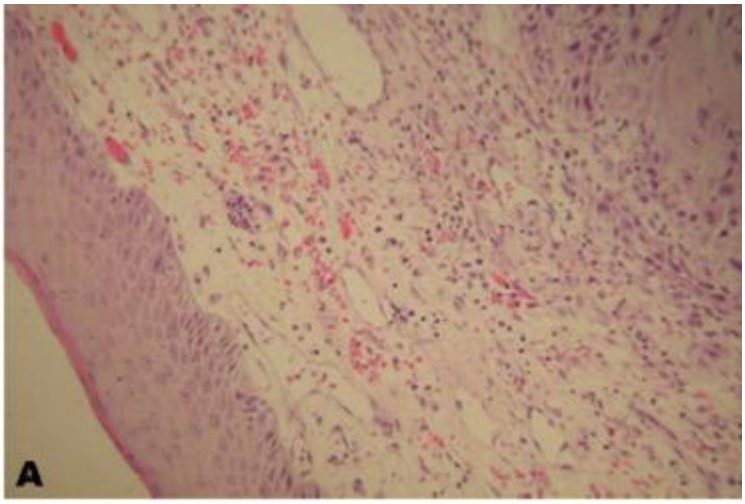

Figure 4. Photomicrographs of the specimen showing A. inflammatory infiltration of the cystic wall (H&E x 10)

Table 4. Histopathological of thyroglossal duct cysts.

Type of cyst	Presumed etiology	Lining epithelium	Stroma features	Mean Location
Thyroglossal duct cyst	Persistence of thyroglossal duct	Stratified squamous, pseudostratified columnar	Thyroid follicles	Midline, hyoid bone

While the current literature reports an equal incidence of thyroglossal duct cysts in males and females, we report a higher incidence in females (71%) which agrees with Shah R. et al. and Yi-Yueh Hsieh, et al. (20,21) and contradictor with the results of many studies [(3)].

A larger population is necessary to determine the true incidence. It is important to note, however, that it has been suggested that a true female predominance does exist amongst familial thyroglossal duct cysts [(5)].

Cysts are usually noted during the first decade of life as a soft tissue mass located on the midline at or immediately adjacent to the hyoid Bone [(22)].

They arise from vestigial remnants of the embryonic thyroglossal duct. Thyroglossal duct cysts had stratified squamous or pseudostratified columnar epithelium with associated thyroid follicles. After reviewing their medical records, we found that these cysts were all located on the midline attached to the hyoid bone. These findings are similar to those of previous Reports [(23,24)].

Considering the embryonic route, they should have been diagnosed as thyroglossal

duct cysts. Thus, proper clinical information should be made available to the pathologist so that a correct diagnosis can be made of these congenital cervical cysts. Thyroid carcinoma may develop in a thyroglossal duct cyst, but its incidence is less than 1 % [13].

Little data exists as to actual incidence of presenting symptoms in patients with TGDC. In our series, 71% of patients had a mass at presentation without infection. In 16% of patients, the neck mass would move with tongue protrusion or swallowing. Tenderness and infection were identified in four patients at a rate of 13%. Of note, Ostlie et al. series showed 22% of patients had preoperative infection [20].

In this series, a history of preoperative infection appeared with almost half the frequency for a total of 13% of our patients. While only 3% of the infants (≤2 years) in this series had preoperative infection, 10% of the school-aged children had preoperative infections. This suggests that older children are more likely to become infected. In our experience, we could not directly correlate age with incidence of preoperative infection; however, a larger study population may show this to be true with statistical significance.

Diagnosis of a TGDC heavily relies on a thorough history and head and neck exam. It is well documented in the literature that MRI, CT, and ultrasound Imaging are all good diagnostic tools for confirming the diagnosis of thyroglossal duct cysts , identification of normal thyroid tissue, and detection of malignancy.

If imaging is necessary, ultrasound Imaging is the ideal initial imaging technique, because it is easily accessible, inexpensive, noninvasive, and does not involve radiation or sedation, which is particularly important in the pediatric population [14].

Conclusions

Mass and infection are the most common presenting symptoms of thyroglossal duct cysts. The incidence of preoperative infection was 13% in our series, which similar to that previously reported [20].It is suggested that school-aged children are more likely to have preoperative infections than younger children.

Complete and precise clinical information is a prerequisite in order for pathologists to make accu

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