DOI: 10.7860/JCDR/2015/14190.6969

Original Article

Surgery Section

Diagnostic and Surgical Approach of Thyroglossal Duct Cyst in Children: Ten Years Data Review

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ABSTRACT

Introduction: Thyroglossal duct cysts are the most common congenital midline neck swelling in children.

Aim: To evaluate the clinical features, treatment, incidence of complications and outcomes in children with congenital midline neck lesions and more specifically with thyroglossal duct cyst, treated in our department the last ten years. It is a retrospective study.

Materials and Methods: The aggregated data revealed 59 patients with congenital midline neck lesions, of which 33 patients were with thyroglossal duct cyst (TDC). The diagnosis of TDC was made by physical examination, ultrasound (US) in all cases, and for complicated cases a magnetic resonance imaging (MRI) was performed. In all cases followed histopathological conformation of thyroglossal duct cyst.

Results: Fifty nine patients were admitted with midline cystic neck mass and the histopathological evaluation revealed, 33 cases (55.9%) of thyroglossal duct cyst, 14 cases (23.7%) of dermoid cysts, 3 cases (5.1%) of second branchial anomalies, 4 cases (6.7%) of inflammation of unknown cause, 4 cases (6.7%) of lymph nodes and 1 case (1.7%) of capillary skin haemangioma. More specifically, 38 patients were admitted with preoperative diagnosis of thyroglossal duct cyst, but in 5 patients pathological investigation demonstrated the presence of dermoid cyst. The mean age of the 33 patients with TDC at the time of surgery was 6.125 years, ranging between 9 months and 13 years with 10 patients younger than 3 years. Of the 38 patients with initial diagnosis of thyroglossal duct cyst, only four patients (10.5%) had a simple cyst excision and complication rate was 25% (1 case with recurrence). In these patients, the histopathological examination showed that three of them had a dermoid cyst and one had thyroglossal duct cyst. The other 34 patients (89.4%) were treated by Sistrunk's procedure, with two cases (5.9%) were proven on histology to be non TDC. In this group the complication rate was 3.03% (1 case with wound infection).

Conclusion: The inappropriate surgical approach due to misdiagnosis or the incomplete surgical procedure remains the impact factor for recurrence. The modification of Sistrunk's procedure remains the main surgical technique that can provides good results with low rate of complication (3.03%) and recurrence (0%) as shown of our collecting data.

Keywords: Congenital, Neck, Sistrunk

INTRODUCTION

Thyroglossal duct cysts (TDC) are the most common congenital midline neck masses in the paediatric population, and account more than 55% of such disorders [1]; the majority appearing before 5 years of age [1,2] with male preponderance. The classic presentation is a midline, non tender, palpable mass that moves with swallowing. Additional presenting symptoms are, midline painless mass (35.7%) midline mass fluctuating in size (30%), dysphagia (12.8%), infected fistula (10%), mid-neck tenderness (8.5%), cough (2.8%) [3]. The differential diagnosis includes ectopic thyroid, pyramidal lobe of thyroid, dermoid or sebaceous cyst, lymphadenitis, thyroid goitre and lipoma [4]. The cysts are ectodermal remnants, and there are several indications for their excision, including recurrent infections, sinus formation, risk of malignancy and cosmetic concerns.

Thyroglossal duct cysts may develop anywhere along the line of embryological descent of the thyroid gland in the neck. Therefore, knowledge of the embryogenesis of the thyroid gland is critical to the successful management of thyroglossal duct cysts. Between the fourth and seventh weeks of embryological development, the thyroid gland descends from the foramen cecum at the base of the tongue along the midline to its final position in the anterior neck. During this time, the hyoid bone also develops from the second branchial arch, and therefore, the thyroglossal duct becomes intimately related to the central portion of the hyoid bone. Under normal circumstances, the thyroglossal duct is obliterated; however, its persistence gives rise to the development of cysts and sinuses [3,5].

The cyst is usually encountered in the thyrohyoid location (60%) within 2 cm of the midline; other locations seen less frequently include submental (24%), suprasternal (13%), and intralingual (2%) [4]. In contrast to branchial cleft lesions, thyroglossal duct cysts typically do not drain through an external opening unless the cyst has become infected or recurs following resection or incomplete excision [6].

An ultrasound and radioisotope scans investigation is often recommended as a preoperative workup to prove the cystic structure of the pathology and to distinguish an ectopic thyroid tissue [7]. However, the information is limited because it is difficult sometimes to exclude other cystic pathologies such as infected or uninfected dermoid cysts or a lymph node abscess.

Excision of the hyoid body and cyst was first proposed by Schlange in 1893 (recurrence rate 20%), but in 1920, Sistrunk first outlined the definitive procedure for thyroglossal duct cyst excision including the removal of the central portion of the hyoid bone and the duct extending to the base of the tongue [2,5,8]. Recurrence rates with Sistrunk's procedure, when properly performed, are less than 3%, and usually present within one year following excision [8,9].

The purpose of the present study is to report our 10 year experience in TDC management emphasizing in well targeted preoperative diagnosis and complications rate and to compare our results with those of other authors.

MATERIALS AND METHODS

We carried out a retrospective review of 59 cases of midline cystic mass that were surgically treated at the 2nd Department of Paediatric Surgery of Aristotle University of Thessaloniki between May 2004 and May 2014. This review revealed 33 consecutive pathologically confirmed cases of thyroglossal duct cysts that underwent a Sistrunk's and non-Sistrunk's procedure. The collected data included: gender, patient's age, length of history and clinical presentation, preoperative investigations, choice of surgical intervention, postoperative complications and postoperative care (follow up) of patients.

RESULTS

Fifty nine patients were admitted with midline cystic neck mass and the histopathological evaluation revealed, 33 cases (55.9%) of thyroglossal duct cyst, 14 cases (23.7%) of dermoid cysts, 3 cases (5.1%) of second branchial anomalies, 4 cases (6.7%) of inflammation of unknown cause, 4 cases (6.7%) of lymph nodes and 1 case (1.7%) of capillary skin haemangioma.

More specifically, 38 patients were admitted with preoperative diagnosis of thyroglossal duct cyst, but in 5 patients pathological investigation demonstrated the presence of dermoid cyst.

The mean age of the 33 patients with TDC at the time of surgery was 6.125 years, ranging between 9 months and 13 years with 10 patients younger than 3 years. There were 22 boys and 16 girls (1.375 male preponderance). All the patients were primary referrals. The length of history before admission ranged from 2 months to 3 years.

Twenty-one patients (63.6%) were admitted with palpable midline cystic mass, eight patients (24.2%) with typical glairy discharge and four patients (12.12%) with serious cervical infection at the midline requiring preoperative drainage and antimicrobial therapy.

The lesions were localized in the midline adjacent to the hyoid bone in 29 cases (88%), above the hyoid in 4 cases (12.12%).

In all patients, ultrasonographic examination revealed unilocular lesions with sonographic appearances ranging from a typical anechoic cyst to a pseudo solid appearance with thin walls and posterior acoustic enhancement. Most of our cases were found to have hyper echoic pattern findings. Even at five patients with dermoid cyst, the ultrasonographic findings were consistent with the presence of thyroglossal duct cyst. The sensitivity of ultrasound examination was 84.22%. Magnetic Resonance Imaging was carried out in four cases with previous infection and in three cases with painful palpable midline cystic mass. The exam demonstrated the presence of TGDC with T1 and T2 high signal. In two patients a radioisotope scan was performed complementary because there was a suspicion of ectopic thyroid gland in ultrasound exam, which finally was not confirmed.

Of the 38 patients with initial diagnosis of thyroglossal duct cyst, only four patients (10.5%) had a simple cyst excision. In these patients the histopathological examination showed that three of them had a dermoid cyst and one had thyroglossal duct cyst. The other 34 patients (89.4%) were treated by Sistrunk's procedure, with two cases (5.9%) were proven on histology to be non TDC. The decision to perform a non-Sistrunk's procedure in the above four cases was taken intraoperatively, because the cyst was separated (in 3 of cases) and there was no continuity of the thyroglossal duct beyond the hyoid bone.

Of 33 patients that have a complete Sistrunk's procedure with a confirmed TDC on histology, suffered postoperative complications only one patient (3.03%) with postoperative wound infection at 8th day after operation. Also, must be noted that there was no complication in the group of patients in age younger than three-year-old. In all patients with Sistrunk operation were placed a penrose drainage, which removed the 2th day after the operation. As far as concerned the group with non-Sistrunk's procedure, there was one patient (25%) with complication of recurrence after 8 months, due to misdiagnoses. The time of hospital stay ranged between 2-10 days. Follow-up periods ranged from 6 months to 4 years.

All data collected are shown in [Table/Fig-1]. The details of one patient (25%) with complication of recurrence after 8 months, due to misdiagnoses are shown in [Table/Fig-2]. The details of four patients (12.12%) with serious cervical infection at the midline requiring preoperative drainage and antimicrobial therapy are shown in [Table/Fig-3].

Total patients with initial diagnosis of TGDC	38
Histologically confirmed TGDC	33
Misdiagnoses based on U/S and confirmed postoperatively	6 (15.75%)
Mean age at surgery (years)	6.125
Gender- male/female	1.375
Length of symptomatology (months-years)	1,5 (2m – 3y)
Sensitivity of Ultrasound and MRI	84.22% - 100%
Preoperative infection	4 (12.12%)
Sistrunk/Non-Sistrunk procedure	34/4
Wound infection	1 (3,03%)
Recurrence (Non-Sistrunk procedure)	1 (25%)
Mean time of recurrence (months)	8 months

[Table/Fig-1]: Collected data.

Age at surgery (years)	Gender	Pre-operative diagnosis	Operation	Histo-pathology findings	Time of recurrence	Re-operation
5	M	Dermoid cyst in U/S	Cyst excision	TGDC	After 8months	No data

[Table/Fig-2]: Data of one case with recurrence

Age	Gender	Incision-drainage + antimicrobial therapy	Time of final operation for TGDC	Operation	Complication
9 months	F	yes	After 4 months	Sistrunk	
13 years	F	yes	After 1 month	>>	
13 years	F	yes	After 1 month	>>	Wound infection
5 years	M	yes	After 2 months	>>	

[Table/Fig-3]: Data of four cases with infected TDC

DISCUSSION

The mean age of clinical presentation in our cases was 6.125 years. There were 22 boys and 16 girls (1.375 male preponderance), which proves that males were affected more frequently compared to females in our study. This fact is in agreement with other studies [5].

Most TDC, are usually encountered in the thyrohyoid location (60%) within 2 cm of the midline; other locations seen less frequently include submental (24%), suprasternal (13%), and intralingual (2%) [4]. In our study the lesions were localized in the midline adjacent to the hyoid bone in 29 cases (88%) and above the hyoid in 4 cases (12.12%).

There is a limited data as to the incidence of symptoms in patients with TDC, but a retrospective review by Josephson and Spencer [3], report that 65.7% of the patients presented with a mass or recurrent mass. In our study that percent of patients was 63.6%. According to our data, there was a clinical suspicion of complicated TDC existence as is apparent from the four cases of cervical infection and the three cases with painful palpable midline cystic mass in which a further investigation was performed.

As far as concerned the preoperative diagnosis, an ultrasonographic examination usually reveals unilocular lesions with sonographic appearances ranging from a typical anechoic cyst to a pseudo solid appearance with thin walls and posterior acoustic enhancement [10]. Most of our cases were found to have hyper echoic pattern findings. The ultrasound helped us to distinguish an ectopic thyroid tissue from a TDC. This issue is very important in children where removal of ectopic thyroid gland that may be the only functioning thyroid tissue could lead to postoperative hypothyroidism. The incidence of ectopic thyroid tissue, in the absence of a normally positioned thyroid ranges between 1-2% [11]. In our study all the patients underwent an ultrasonography with rate sensitivity touches 84.22%. None of the patients had an ectopic thyroid gland in ultrasound imaging. In seven patients a MRI was carried out in order to have a detailed illustration of the position, hyoid bone attachment and extension of TDC. However, ultrasound is difficult sometimes to exclude other cystic pathologies

such as infected or uninfected dermoid cysts or a lymph node abscess. Especially dermoid cysts in the midline are often clinically misdiagnosed as TDC [12]. In our study 5 cases proved hystologically to be dermoid cysts even though we had preoperative findings of TDC. We prefer ultrasound as method of choice to determine the presence of normal thyroid tissue due to its low cost and availability in children. We propose that complementary imaging may be used only in selected and complicated cases.

Thyroglossal cyst excision as described by Sistrunk in 1920 always includes excision of the suprahyoid thyroglossal duct until the level of the foramen caecum at the base of the tongue, to prevent recurrences [13]. The rate of recurrence was 3% in Sistrunk's first series as reported by Bennett et al., [14]. Recurrence was acceptable at 3.7% in this series. In our study we had a recurrence rate 0%.

In our days many surgeons do not perform the original Sistrunk's procedure and stop short of the foramen caecum to avoid breaching the pharynx [8,15]. This modification of the Sistrunk's procedure remains the treatment of choice for the thryroglossal duct cysts up to date. In all 34 cases we performed this concrete surgical technique with an excision including the removal of the central portion of the hyoid bone, the duct and a core of tissue around him, extending to the base of the tongue.

Attention has been drawn to the fact that the thyroglossal duct frequently branches and forms duplications around the hyoid bone without the surgeon having chance to recognize all of them [16]. This fact provides the opportunity to the cyst to recur if enough tissue surrounding the main duct is not excised [17]. For this reason Horisawa et al., have recommended the removal of a core from the base of the tongue of approximately 5mm in depth, in order to encompass the foramen caecum [16].

Other factors, which are responsible for recurrences are; young patient age, lobulation of the cyst, rupture of the cyst during operation, inflammation and/or infection and patients with fistulas [18]. In contrast there are suggestions that inappropriate surgical approach intraoperatively or due to initial misdiagnosis is the reason for recurrence [13]. In our study we had 4 cases with initial midline cervical infection in which a preoperative drainage and antibiotic therapy was performed [Table/Fig-3]. As collecting data shows no recurrence mentioned. In one case there was only a wound infection, which prolongs patient's hospital stay. In our opinion, the factor that has the greatest impact for recurrence, is the wrong surgical approach due to misdiagnosis as demonstrated in our case (e.g. simple cyst excision in one patient with dermoid cyst attached indirectly to the hyoid bone), or incomplete surgical procedure. We do not consider the patient young's age to be a factor in recurrence if the principles of the surgical procedure are strictly followed. In our study there was no recurrence among the ten patients younger than three-year-old. In addition our findings suggest that appropriate control of the patients with prior infection may be the key to avoid any kind of postoperative complication. We think that pre- and postoperative antimicrobial therapy, timing of surgery after resolution of inflammation and extensive and radical surgical approach are catalytically correct response points.

In this point we must pay a special attention to the dermoid cysts, which diagnosed during the operation. DeMello et al have proposed that the cysts represent variants of thyroglossal duct cysts with predominating ectodermal tissue and performed Sistrunk's procedure for all dermoid cysts attached to the hyoid bone [12]. For this reason we believe that Sistrunk's procedure should be done for dermoid cysts attached to the hyoid bone. Recurrence rates with Sistrunk's procedure, when properly performed, are less than 3%, and usually present within one year following excision [8,9]. Most studies note recurrence in approximately 2-3% of patients following a Sistrunk procedure [19,20].

CONCLUSION

We can conclude that the combination of clinical and radiological assessment consist a necessary condition for the improvement of the diagnosis sensitivity's. The inappropriate surgical approach due to misdiagnosis or the incomplete surgical procedure remains the impact factor for recurrence. The appropriate management of the patients with prior infection may be the key to avoid any kind of postoperative complication. The young age of patient does not appear to play a role in recurrence rate. The modification of Sistrunk's procedure remains the main surgical technique that can provide good results with low rate of complication (3.03%) and recurrence (0%) as shown in our collecting data.

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FINANCIAL OR OTHER COMPETING INTERESTS: None.

Date of Submission: **Mar 27, 2015**
Date of Peer Review: **Jun 08, 2015**
Date of Acceptance: **Jul 06, 2015**
Date of Publishing: **Dec 01, 2015**