

ORIGINAL ARTICLE**Management of Thyroglossal Duct Remnants: Our Experience**

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Abstract

Background: Thyroglossal duct remnants are more common in paediatric population. Complete excision of the tract is necessary to prevent recurrence. Sistrunk's surgery is done for thyroglossal duct remnants. This surgery has considerable failure rate. **Aims and Objectives:** This study was done to present our clinical experience regarding the successful management of thyroglossal cyst and fistula to know the role of imaging in prevention of recurrence after Sistrunk's surgery. **Material and Methods:** This is an observational study of 15 cases of thyroglossal cyst and fistula. The data was collected retrospectively from the medical records of SDM College of Medical Sciences and Hospital. **Results:** No recurrence was seen in this series with a follow up period of 1 year. **Conclusion:** CT fistulography for thyroglossal fistula and USG neck for thyroglossal cyst is the imaging modality of choice. Nevertheless, it is pivotal to excise a wider core of tissue in revision cases to ensure complete cure to prevent possible recurrence.

Keywords: Cyst, Fistulography, Hyoid, Sistrunk's, Thyroglossal

Introduction:

Anomalies of the thyroglossal duct are seen commonly in the pediatric population; they are second in frequency only to lymphadenopathy. Remnants of the thyroglossal duct are actually found in approximately 7% of the population, but only a small number of these ever become symptomatic. Thyroglossal duct remnant may present as a cyst, fistula or abscess. This congenital swelling is excised by Sistrunk's operation and are amenable to complete cure. As the tract can have multiple

branches it is prone for recurrence if not completely excised. There is a high recurrence rate reported particularly in revision cases. No recurrence is seen in our series as wider core of tissue excision done after appropriate pre-operative imaging.

Material and Methods:

The study was done in the department of ENT in a tertiary health care centre. A total of 15 cases of Thyroglossal Duct Remnants (TGDR) were studied of which 9 were Thyroglossal Cyst (TGC) and 6 were Thyroglossal Fistula (TGF). All cases who presented with TGC which was confirmed by Ultrasound and Fine Needle Aspiration Cytology (FNAC) and patients with thyroglossal fistula confirmed by Computed Tomography (CT) Fistulography were included in the study. There were no exclusion criteria. Data regarding the history, operation, investigations and follow up were collected by reviewing the case records obtained from the Medical Records Department of a tertiary care center.

All the cases were operated with wide core tissue excision and followed up during the time period from January 2008 to January 2013.

Results:

The study group consisted of 15 patients of which 9 patients were TGC and 6 were TGF. There were 6 pediatric cases and 4 female adult patients. Most patients presented in the 2nd decade of life. Out of 9 TGC two patients had suprahyoid and 7 had infrahyoid position. Out of 6 TGF cases, 5 cases

were as a result of incomplete excision in other centers and one was a result of spontaneous rupture of cyst. One of TGF patient had undergone 2 revision surgeries. Two patients had suprahyoid and four had infrahyoid TGF.

Pie chart and bar diagram (Fig.1 & 2) shows the age distribution of the cases and the site of TGDR in relation to hyoid respectively.

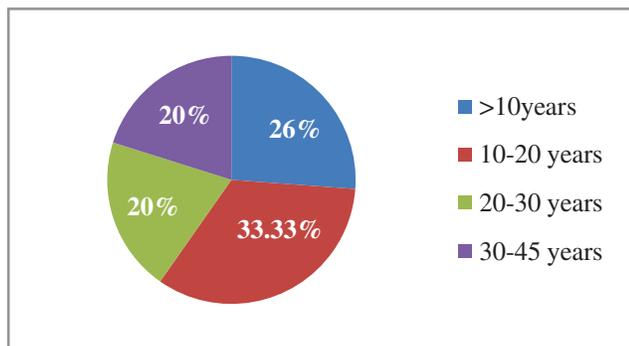


Fig.1: Pie Chart Showing Age Distribution of TGDR Cases

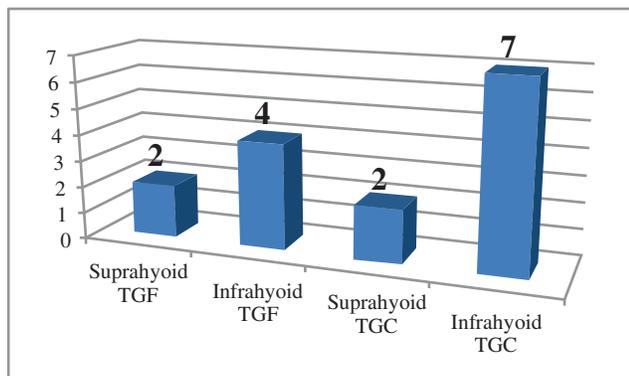


Fig. 2: Bar Diagram Showing Number of Suprahyoid and Infrahyoid TGC and TGF

Five of the TGF cases were a result of incomplete excision in other centers and one was a result of spontaneous rupture of cyst. One of TGF patients had undergone 2 revision surgeries. All the TGC (Fig.3) patients were confirmed by Ultrasonography (USG) and FNAC and all the TGF patients were subjected to CT Fistulography.

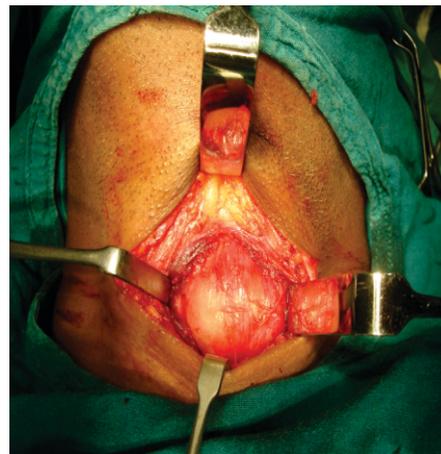


Fig 3: Intraoperative Photograph of TGC

In two of them the tract had undergone significant scarring due to previous surgery and hence the tract was not patent (Fig.4).



Fig. 4: CT Fistulography Showing Tract Relation to Hyoid Bone

An intravenous cannula was used to introduce dye into the tract. In one of the cases the tract was trifurcated within the tongue with the central tract ending at the foramen caecum (Fig.5 & 6) and the other two branches ending abruptly within the tongue. Accessory tracts to the main tract were present in about 8% of the cases [1].



Fig 5: CT Fistulography Showing Tract at Base of the Tongue

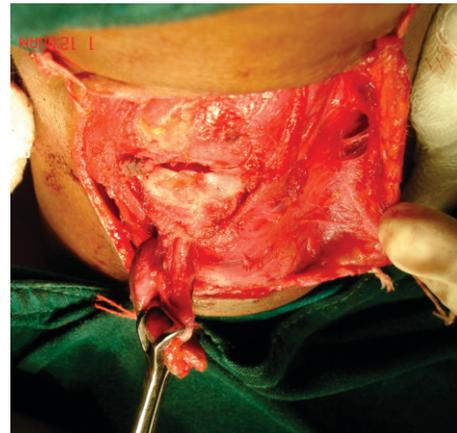


Fig.7: Intraoperative Photograph of TGF



Fig. 6: Sagittal Section of CT Fistulography Showing the Entire Course of the Tract

TSH levels have been ascertained in all the patients irrespective of the position of the cyst. All the patients were treated with preoperative antibiotics empirically without being subjected to aspiration or incision and drainage to avoid spillage of the cyst content and risk of recurrence.



Fig. 8: Excised Fistula Tract

Sistrunk's operation (Fig.7 & 8) was done for all patients taking a wider core of tissue for all the recurrence cases. The post operative follow up of patients ranged from 6 months to 1 year. None of the patients had recurrence. Recurrence was defined as persistent neck mass or fistula following surgery. The success rate was 100 % in this series as opposed to other studies with 20% to 35% recurrence following revision surgery [2]. The only complication noted in this series is hypo pigmentation at the scar site following surgery in two patients.

Discussion:

The thyroid gland is the earliest endocrine derivatives of the pharynx to develop. It originates from the foramen caecum at the floor of the pharyngeal gut on day 17 of gestational development as a single midline evagination of the endoderm. The diverticulum then grows caudally in a loose subpharyngeal plane of mesoderm and also proliferates at its tip into two lateral lobes. This connection tracks anterior to the hyoid bone, reaches the normal position by about seventh week of embryonic life and the duct usually disappears by the tenth week. The remnants may persist in the form of cyst, fistula, sinus or abscess. The relationship of the thyroglossal tract to the body of the hyoid is important. The thyroglossal tract starts from the pyramidal lobe of thyroid, ascends on the anterior surface of the thyrohyoid membrane, curves along the posterior surface of the hyoid, ascends along the anterior surface of hyoid bone and ends in base of the tongue. The pyramidal lobe indicates its former site of attachment to the thyroid gland. It is essential to excise the tract throughout its entire length from the foramen caecum to the pyramidal lobe along with the middle segment of body of hyoid to prevent recurrence.

The prevalence rate of TGDR is 7 % with a 25 % off the midline presentation [3]. TGCs that occur off the midline may be difficult to differentiate from branchial cleft cysts. Although a congenital swelling, only 50 % present in the first decade and up to 66 % present within the 3rd decade of life [4]. In 2/3rd of the patients they are found immediately adjacent to the body of hyoid. Other sites include between the tongue and hyoid, between the hyoid and pyramidal lobe of thyroid, within the tongue, or within the thyroid. Due to attachments to the base of tongue, TGDRs move superiorly in the neck when the patient protrudes the tongue or

swallows. Adults usually present with an infected cyst or abscess presenting as pain and redness of swelling. 25 % present with fistula which may result from spontaneous or surgical drainage [2]. Leakage of saliva may be noted through the opening. Drainage into mouth through foramen caecum may produce halitosis or dysphagia. Cyst at the base of tongue is a rare and can present with stridor in a neonate.

Sistrunk's operation is the definitive treatment for thyroglossal duct remnants which involves complete excision of the cyst or fistula and its embryonic tract to avoid recurrence. The pyramidal lobe of the thyroid, the thyroglossal duct cyst or fistula, the central portion of the body of the hyoid bone and a broad core (≥ 1 cm) of suprahyoid muscle extending up to the foramen caecum has to be excised. The body of hyoid bone must be excised due to the inconsistent nature of the tract in relation to it. Recurrence is almost always a result of incomplete excision and can be present as persistent neck mass or a draining fistula. Recurrence of TGC after complete excision using the Sistrunk's procedure is reported to be 2.6 % to 5 % whereas simple excision of cyst can result in recurrence rates as high as 38 % to 70 % [4, 5]. Brousseau VJ *et al* have reported a recurrence in 2 cases in a series of 62 patients comparing the result of Sistrunk's surgery in adults and children [6]. Ein S H *et al* has reported a recurrence rate of 10 % in a series of 270 patients with most recurrences occurring when the middle third of hyoid is left intact [7]. Athow AC *et al* have concluded in their series of 40 children with suspected thyroglossal cyst that it is better to perform early Sistrunk's operation in any lesion whose presentation is typical of thyroglossal cyst [8]. Shah R *et al* have reported a recurrence rate of 3.4 % in their series of 29 patients who underwent Sistrunk's procedure [9]. Recurrence rates ranging from 1 %

to 30 % have been reported in few other series [10-12]. The most common cause of recurrence is rupture of the cyst intra-operatively or leaving part of the wall behind. A dumbbell shaped cyst deep in the back of the hyoid bone pushing the thyrohyoid membrane posteriorly can be easily missed if appropriate imaging is not done. A cyst that has ruptured to form thin walled pseudo cysts is also difficult to excise completely. Amateur surgeons often fail to realize that the tract may be multiple and take too thin a core which often leads to recurrence. They apprehend the difficulty in speech and swallowing and hence leave behind suprahyoid portion of the tract adjacent to the foramen caecum. Perkins JA *et al* have attributed 20 % recurrence to lack of base of tongue musculature removal [13]. Perioperative infection has been proposed as a cause of recurrence in few studies [14]. Recurrent TGCs are more difficult to deal with due to distorted anatomy, loss of fascial planes as a result of fibrosis. Hence the recurrence is higher following surgery for recurrent TGDRs. Mikel RA *et al* have reported success in recurrent cases with en bloc anterior neck dissection which is a modification of classical Sistrunk's procedure [15].

Recurrence can be avoided by a preoperative course of antibiotic to eradicate infection. Needle aspiration decompresses the cyst and culture and sensitivity allows appropriate antibiotic coverage for infection. Occasionally the cyst contains all of the functioning thyroid tissue. Because of the potential for permanent hypothyroidism after surgical excision, many investigators advocate routine preoperative assessment of the thyroid. Absence of the thyroid gland at the level of the thyroid cartilage should redirect attention to the tongue for an ectopic lingual thyroid gland. An ultrasound helps to rule out ectopic thyroid which is rare (1-2 %) but if present is the only thyroid

tissue in 75 % of patients [16]. It also provides information regarding consistency of the cystic lesion. Tc99m scans provide functional information regarding thyroid tissue. They are useful to rule out ectopic thyroid and to determine the presence of normal functioning thyroid tissue before any consideration for surgical excision but may not be available in many centers. Fluoroscopic Fistulography delineates the tract in a TGF but it does not provide cross-sectional details. CT scan is the workhorse imaging modality for adult neck masses. It provides three-dimensional relationships, excellent details of mucosal disease and involvement of adjacent bone. MRI offers superior soft tissue delineation. Recurrent TGDR management becomes challenging because cysts may be multifocal with the presence of fibrosis, distorted surgical landmarks and possible absence of hyoid bone. Both CT and MRI provide good cross-sectional anatomical details but fail to delineate the course of the fistulous tract and its patency. CT Fistulography provides a panoramic view combining the advantages of both the modalities. Excision of a wide core of tissue above the hyoid bone is the key to reduce postoperative recurrence [17].

Conclusion:

Thyroglossal duct remnants are treated with Sistrunk's procedure. The recurrence rate following surgery is quite high and can be avoided by appropriate imaging prior to surgery. Ultrasound is adequate for imaging TGC and surgeon has to take a wide core of tissue along the tract to avoid recurrence. TGF which is usually the result of incomplete excision is more difficult to treat and CT Fistulography is the best modality to delineate the tract particularly in cases where the tract is multiple and ensures complete excision.

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