Management of Thyroglossal Fistula and Recurrent Thyroglossal Duct Remnants: A New Concept for Surgical Approach

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Abstract

Background: Surgery for thyroglossal duct cysts has a high cure rate, however, surgery for recurrent cases (thyroglossal fistulas) has less success rate and there are still recurrences despite a competently performed procedure. In addition, many patients who present with thyroglossal fistula without history of previous surgery have lower cure rates. All these patients present a surgical challenge since no standard treatment exists. Surgery for such cases may encounter some difficulties due to abnormal anatomy as a result of previous surgery or infection leading to fibrosis and adhesions in addition to the unexpected branching pattern of the tract. The management of the thyroglossal fistula could be as a pharyngeo-cutaneous fistula management with tight pharyngeal closure after proper excision of the identified tract with the central hyoid (if not excised before) and to avoid wide area of dissection and excision which has a considerable risk of complications.

Aim of the Work: To evaluate and discuss the outcome of tight pharyngeal closure by hyoid gap closure and layered closure in the management of thyroglossal fistula and recurrent thyroglossal duct remnants.

Methods: A retrospective study conducted from March 2014 to January 2016 including 27 patients suffering from thyroglossal fistula having 16 patients as a primary presentation without surgical history and 11 patients of recurrent disease after Sistrunk operation for thyroglossal cyst. All patients have been presented and managed at the Department of Otolaryngology, Hearing and Speech Institute and El-Sheihk Zayed Specialized Hospital. All patients have undergone tight pharyngeal closure by hyoid gap closure and layered closure. Regular follow-up visits were done every two months for at least one year.

Results: The operative time ranged from 1 to 2 hours with a mean of 1.2 hours. Blood loss has a range of 55-170 ccms with a mean of 90 ccms. Hospitalization was one day in all patients with discharge in the second postoperative day. As regards complications, there were no major complications in the form of injury to important adjacent structures or massive hemorrhage. Other complications in the form of wound infec-

tion in one patient was evident 1 week after surgery and well controlled by parental antibiotics and local antiseptic measures. Return to normal life activity or work was possible in all patients after 10 days. As regards the final outcome, all patients were doing well throughout the follow-up period. There was no recurrence in 26 patients of the study except one male patient who suffered recurrence of discharging fistula after nine months of our surgery. In this case the recurrence was most probably due to infrahyoid remnants. This patient was re operated by central neck dissection after 11 months of the first surgery and doing well until the time of writing.

Conclusion: Tight hyoid closure and layered closure have successfully managed thyroglossal fistula and recurrent thyroglossal duct remnants. Understanding the anatomy and pathophysiology is important in treating these challenging cases.

Key Words: Recurrent thyroglossal fistula – Hyoid closure – Pharyngeal fistula.

Introduction

THYROGLOSSAL cyst disease is the most common developmental neck lesion in the pediatric group. There is a slight male predominance. These lesions usually present as a midline mass, but can present initially as a draining sinus or abscess [1,2]. Most thyroglossal duct cysts present during the first 5 years of life, although the lesion has been repeatedly described during adulthood [3]. Associated ectopic thyroid tissue was found in one case specimen in a study, but thyroid gland was in normal place [4]. An author studied the recurrence rate after Sistrunk operation and reported a case with an intrathyroidal thyroglossal cyst [4].

Subsequently, the removal of a thyroglossal duct cyst is a commonly performed surgical procedure in children. The successful management of this midline neck mass depends on a thorough understanding of the embryogenesis of the thyroid gland and the thyroglossal tract [5].
The thyroid gland begins its early development at the level of the foramen cecum of the tongue base. Normally, the thyroglossal duct descends as a bilobed thyroglossal diverticulum to form the normal thyroid gland in the anteroinferior neck region during embryogenesis. During the descent, the tract passes through the midline tongue musculature and comes into close relationship with the hyoid bone Fig. (1). The thyroglossal duct, which connects the thyroid gland with the foramen cecum, normally involutes from inferior to superior fashion. Failed involution of the thyroglossal duct can lead to the formation of thyroglossal duct cysts [6].

Historically, extremely high recurrence rates were reported with incision and drainage, and simple cyst excision procedures. The understanding and appreciation of the embryology and pathophysiology of thyroglossal duct cysts lead to the belief that persistent thyroglossal duct remnants were mainly due to incomplete excision at the level of the hyoid bone and beyond. In 1893, Schlange described a procedure that involved the excision of the mid-portion of the hyoid bone in continuity with the main neck mass [7]. Later in 1920 and 1928, Sistrunk added the removal of additional core of tissue between the hyoid bone and the foramen cecum, theoretically resulting in a complete eradication of the thyroglossal tract [8,9].

Sistrunk’s principles of surgical management of thyroglossal duct cysts has largely been accepted and is currently the standard of treatment. Moreover, there is a large body of literature that supports the Sistrunk operation with excellent results [10]. Although resulting in a substantial reduction in the postoperative recurrence rates, many cases of thyroglossal duct cysts continue to recur despite the practice of seemingly adequate surgical resection [11].

The role of infection has been evaluated in many studies. In a study, only one patient (5%) had a history of infection and the culture showed Homophiles influenza [12]. In more recent study, preoperative infection was detected in 10 cases: Seven cases were of cellulitis and three cases were of abscess, which was treated with incision and drainage. In this study, there were seven recurrent cases and most of them were due to postoperative infection. Infection may cause rupture of the cyst and implantation of the epithelium of the cyst into the surrounding tissue and is responsible for recurrence [13]. Recurrence rate of thyroglossal cyst after complete excision using the Sistrunk procedure is reported to be 2.6-5%, whereas simple excision of the cyst can result in recurrence rates as high as 38-70%. Other authors [14,15] have reported a recurrence in two cases in a series of 62 patients. Ein et al., [16] reported a recurrence rate of 10% in a series of 270 patients, with most recurrences occurring when the middle third of the hyoid was left intact.

Authors [17,18] have reported a recurrence rate of 3.4% in their series of 29 patients who underwent the Sistrunk procedure. Recurrence rates ranging from 1 to 30% have been reported in a few other series [19,20]. The most common cause of recurrence is rupture of the cyst intraoperatively or leaving a part of the wall behind.

For a thyroglossal duct fistula persisting after a Sistrunk procedure, a number of solutions have been reported in the literature with the goal of complete excision of the thyroglossal duct remnant from the thyroid gland to the tongue base region [21]. Mainly, these methods involve en bloc tissue removal to fully encompass the variable thyroglossal duct pathways but there remains a debate about how much and which tissue needs to be removed, resulting in a variety of procedures being reported with varying success rates [22].

There are three cornerstone considerations before starting my work and to conclude the new concept of surgery. The first, is the fact of being a fistula not a sinus, so it should connect the pharynx with the exterior. With good understanding of the embryology and anatomy, we know that the pharyngeal end lies at the foramen cecum of the tongue base. The second consideration is to manage this fistula as a pharyngocutaneous fistula aiming to do tight pharyngeal closure after proper excision of the identified tract with the central hyoid (if not excised before) and even if we bury epithelial remnant or branches under the tight closure [23]. The third consideration is based on the histopathological and imaging proof of the site of recurrence to be suprathyroid or retrohyoid segment in most or all cases and the highly variable branching pattern mandating wide area of dissection and excision to avoid recurrence and this carry more risk of complications which we are aiming to avoid in our study [24].

The objective of this study is to evaluate and discuss the outcome of tight pharyngeal closure by hyoid gap closure and layered closure in the management thyroglossal fistula and recurrent thyroglossal duct remnants.

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**References**


[13] The objective of this study is to evaluate and discuss the outcome of tight pharyngeal closure by hyoid gap closure and layered closure in the management thyroglossal fistula and recurrent thyroglossal duct remnants.
Subjects and Methods

Ethics:
We have obtained a written informed consent from each patient accepting to participate in the study according to and approved by the Institutional Ethical Committee.

Subjects:
This is a retrospective study conducted from March 2014 to January 2016 including 27 outpatients at the Department of Otolaryngology, Hearing and Speech Institute and El-Sheikh Zayed Specialized Hospital with a clinical diagnosis of thyroglossal fistula or recurrent thyroglossal remnants. We selected outpatients (19 males, 8 females) with discharging upper cervical midline orifice. The discharge was scanty intermittent mucoid in all patients except in two patients who presented with mucopurulent discharge. 16 patients presented with the primary pathology without history of surgery, however 11 patients presented to us with recurrent disease having the fistula in the scar of the previous surgery. All of those patients, aged from 12 years to 38 years (median 24.6 years), had been suffering skin discharge for an average of 3.9 years. All of the patients were followed-up for 22 months. When patients agreed to participate in the study, they were informed that they must provide information about the occurrence of any swelling or skin discharge they had experienced (Table 2).

Methods:
All patients underwent detailed medical and surgical history, complete general and local examination. In addition, the protocol of diagnosis included an examination by CT scanning or MRI to exclude other possible causes of skin fistula especially in recurrent cases.

After the appropriate selection of patients, general anesthesia is inducted via oral endotracheal tube. Small amount of sterilized dye (methylene blue) is injected in the fistula as a trial to delineate the tract during dissection. A horizontal skin incision was done about 2 inches long at or just below the level of the hyoid bone in the midline of the neck. The incision includes the fistula in an elapse and is carried down through the subcutaneous tissue and platyzma muscle. Once the platyzma and skin are elevated, the tract can be visualized and followed down to the hyoid bone. The strap muscles are retracted laterally and the tract is dissected free of the thyroid cartilage and surrounding tissue until it is pedicled superiorly to the hyoid bone. The muscles and soft tissue are then dissected of the central segment of the hyoid bone about 1.5 to 2cms in length avoiding superior and inferior dissection to avoid injury of the tract. The hyoid bone is then cut on each side. A bloc of tissue including the fistula tract and the central hyoid is followed to the supra and retro hyoid directions in the classic way to excise a core of tongue muscles with or without excision of the foramen cecum [25].

The new concept:
Until this step, there was no difference from the standard Sistrunk surgery but, also we are not sure of complete eradication of all epithelial or thyroglossal remnants. Now we perform hyoid closure by approximating both cut ends of the hyoid bone to each other and fixation of them tightly in a vertical line of closure by 1 zero vicryl sutures 3 to 5 in number. So by this technique we close a substantial dead space created by removal of a portion of the hyoid bone and a core of tongue muscles. Again this space may harbor thyroglossal remnants which lose its connection to the subcutaneous space preventing recurrence but still possess its natural connection to the pharynx through the foramen cecum Fig. (2).

Then we re approximate the strap muscles and suture them by vicryl sutures, followed by continuous stitch closure of the subcutaneous layer and skin closure by tight interrupted silk sutures or subcuticular Prolene stitch. So we obtain four layers of tight closure without leaving a drain after proper hemostasis [Fig. (3) & (Table 1)].

The diagrams in Figs. (1-3) were obtained from web sites of anatomy photos, whoever i have modified these photos utilizing the paint program in my P.C. according to the surgical steps to clarify and illustrate the new concept of surgery especially in Figs. (2,3).

In the eleven patients with recurrence after Sistrunk surgery, the situation was different with some difficulties. Identifying tissue planes, and following the tract through the hyoid defect and tongue base was done. After excision of the thyroglossal remnant carrying tissue bloc we again re approximated the two cut ends of the hyoid bone after re freshening or trimming them, then fixed tightly to each other. Then we proceed in the same way mentioned above.

Postoperative care in the form of proper wound dressing and sterilization, parenteral broad spectrum antibiotics for one week followed by oral antibiotics
for another one week. Stitch removal after two weeks of surgery with good inspection of the wound for signs of infection or discharge.

Follow-up data were available for all 15 patients. Follow-up ranged from 1 year to 22 months, with the mean follow-up period of 1.2 years.

Exclusion criteria:

We excluded patients with thyroglossal cysts, lateral cervical sinuses or fistulas, patients with suspected malignancy or post irradiation patients.

Table (1): The new concept of tight four layers closure.

<table>
<thead>
<tr>
<th>Layer</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>First</td>
<td>Hyoid closure by approximating both cut ends of the hyoid bone to each other and fixation of them tightly in a vertical line of closure by 1 zero vicryl sutures 3 to 5 in number in the periostium or attached muscles. So by this technique we close a substantial dead space created by removal of a portion of the hyoid bone and a core of tongue muscles. Again this space may harbor thyroglossal remnants which lose its connection to the subcutaneous space preventing recurrence but still possess its natural connection to the pharynx through the foramen cecum.</td>
</tr>
<tr>
<td>Second</td>
<td>Then we re approximate the strap muscles and suture them by vicryl sutures.</td>
</tr>
<tr>
<td>Third</td>
<td>Continuous stitch closure of the subcutaneous layer.</td>
</tr>
<tr>
<td>Fourth</td>
<td>Skin closure by tight interrupted silk sutures or subcuticular Prolene stitch. Four layers of tight closure without leaving a drain after proper hemostasis.</td>
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</tbody>
</table>

Delayed skin stitch removal (at 14 days postoperative) under umbrella of broad spectrum antibiotics minimizing the chance of recurrence of a pharyngocutaneous fistula.
Results

The operative time ranged from 1 to 2 hours with a mean of 1.2 hours. Blood loss has a range of 55-170ccms with a mean of 90ccms. The operative time was longer in the revision cases. Hospitalization was one day in all patients with discharge in the second postoperative day. As regards complications, there were no major complications in the form of injury to important adjacent structures or massive hemorrhage. Other complications in the form of wound infection in two patients was evident 10 days after surgery and well controlled by parenteral antibiotic and local antiseptic measures. Return to normal life activity or work was possible in all patients after 10 days. Pain was moderate in all patients, but was sever in four patients of recurrence due to marked tissue dissection and trauma (Table 3).

As regards the final outcome, all patients were doing well throughout the postoperative period and the follow-up visits. There was no recurrence in 26 patients of the study. Only one male patient which was presenting by recurrent thyroglossal fistula suffered recurrence of discharging fistula after nine months of our surgery. In this case the recurrence was most probably due to infrahyoid remnants. This patient was re operated by central neck dissection after 11 months of the first surgery and doing well until the time of writing (Table 4).

Table (2): Pathology and sex distribution.

<table>
<thead>
<tr>
<th>Pathology at presentation</th>
<th>Male</th>
<th>Female</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Thyroglossal fistula</td>
<td>12 patients</td>
<td>4 patients</td>
<td>16 patients</td>
</tr>
<tr>
<td>Recurrent thyroglossal fistula</td>
<td>7 patients</td>
<td>4 patients</td>
<td>11 patients</td>
</tr>
<tr>
<td>Total</td>
<td>19 patients</td>
<td>8 patients</td>
<td>27 patients</td>
</tr>
</tbody>
</table>

Table (3): Results and perioperative data.

<table>
<thead>
<tr>
<th>Operative data and outcome</th>
<th>Range</th>
<th>Mean</th>
</tr>
</thead>
<tbody>
<tr>
<td>Operative time</td>
<td>1-2 hours</td>
<td>1.2 hours</td>
</tr>
<tr>
<td>Blood loss</td>
<td>55-170ccms</td>
<td>90ccms</td>
</tr>
<tr>
<td>Age</td>
<td>12-38 years</td>
<td>24.6 years</td>
</tr>
<tr>
<td>Follow up</td>
<td>15-23 months</td>
<td>18 months</td>
</tr>
<tr>
<td>Postoperative hospitalisation</td>
<td>1 day in all patients with discharge at the second postoperative day</td>
<td></td>
</tr>
<tr>
<td>Return to normal life activity</td>
<td>In all patient after 10-14 days</td>
<td></td>
</tr>
<tr>
<td>Complications</td>
<td>Major (injury to important structures, massive hemorrhage)</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Wound infection</td>
<td>2 patients</td>
</tr>
<tr>
<td></td>
<td>Chest infection</td>
<td>-</td>
</tr>
</tbody>
</table>

Table (4): Results (final outcome).

<table>
<thead>
<tr>
<th>Parameter</th>
<th>No recurrence</th>
<th>Recurrence</th>
<th>Timing of recurrence</th>
<th>Action</th>
<th>Percentage</th>
</tr>
</thead>
<tbody>
<tr>
<td>27 patients of the study</td>
<td>26 patients</td>
<td>1 Male patient</td>
<td>9 months after surgery (recurrent thyroglossal fistula) i.e. second recurrence</td>
<td>Midline neck dissection after 11 months of surgery or 2 months of recurrence</td>
<td>3.7%</td>
</tr>
</tbody>
</table>
Discussion

Several authors have demonstrated that the recurrent thyroglossal duct is often due to the multiple, microscopic, irregular, and branching nature of the tract, which cannot always be detected by the surgeon in the operating room. More specifically, several accessory tracts and variable patterns of branching to the tongue base in the suprahypoid region have been reported. For this reason, it can be assumed that the recurrent thyroglossal remnants in its entirety cannot be removed unless a wide core of tissue is excised along the path of the thyroglossal tract [26].

Traditionally, the wider resection approach was advocated by several authors in hopes of removing all of the branching remnants in cases of recurrent thyroglossal remnants. Hoffman et al., reported definitive cure in all recurrent patients with further, wider excisions [27]. Their histological review showed high variability in suprahypoid patterns of drainage, with accessory tracts of the main duct being found in multiple specimens. Moreover, some of the accessory tracts and alveolar outpouchings were intimately associated with the pharyngeal mucosa and the surrounding striated muscles on the tongue (genioglossus and myelohyoid). This further indicates that a core tissue of muscle and mucosa may be required for the recurrent cases [28].

Histological analysis of the specimens showed that there were three cases misdiagnosed preoperatively as thyroglossal cyst, but histopathological examination postoperatively showed two cases of lymph nodes and one case of dermoid cyst. Athow et al., [29] stated that dermoid cysts, second in incidence to thyroglossal duct cyst, are reported to sometimes occur simultaneously with thyroglossal duct cyst.

As mentioned above, many earlier studies focused on the wider excision of the thyroglossal duct to fully excise the multiple and accessory tracts that are present in recurrent lesions. In addition, some authors reported removal of additional hyoid bone at the lateral cut-ends. Several authors, advocated for the complete removal of residual tracts deep to the previously removed hyoid bone and also emphasized the importance of removing the epithelial remnants that deviated laterally from the midline. Similarly, Hoffman also suggested that a wider excision of the suprahypoid tissue up to the level of the pharyngeal mucosa with a margin of tongue base muscle be included in recurrent cases. As well, he recommended that skeletonizing the tract above the hyoid bone should be condemned, since it does not account for the accessory tracts [31].

A review of 38 recurrences in 143 patients by Pelausa et al., showed that inadequate hyoid bone resection and persistent peri-hyoid tract remnants were the most common causes for recurrences. Subsequently, the revision operation included additional hyoid bone removal and suprahypoid core of tissue excision to the foramen cecum and good cure rates were demonstrated afterwards. Moreover, this study recommended that a 0.5cm radius core of tissue from midportion of the hyoid bone to tongue base be removed [32].

In a recent study, prevention of recurrence of thyroglossal cyst and fistula necessitates complete excision of the track and central portion of the hyoid bone. In this study, infection is a very important factor preoperatively and postoperatively that increases the risk of recurrence of thyroglossal fistula. Postoperative infection is associated with a higher rate of recurrence compared with preoperative infection. They recommended Sistrunk operation as an ideal surgery for the treatment of thyroglossal cyst and fistula with every effort taken to prevent postoperative infection as it is the important factor causing recurrence of the disease [13].

In practice, excision of the suprahypoid tissue can be quite challenging, since the tract is ill defined and some have proposed that this area may actually represent a pseudocyst rather than a true thyroglossal duct remnants. There is some controversy regarding the amount of hyoid bone that requires excision to prevent recurrences. Most authors report that the central 2/3 rd of the hyoid bone or the area between the inferior cornuæ must be removed to reduce the risk of leaving tracts in situ but this has not been proven [33].

Some authors have advocated the excision of the foramen cecum itself to prevent further recurrences, while others suggest that entry through the tongue base into the oropharynx is not necessary for definitive cure. One study reported a combined transoral/cervical approach to remove a core of tissue around the foramen cecum for a rare intra-lingual cyst recurrence. Currently, there is no convincing evidence that foramen cecum and a core of tongue base mucosa needs to be excised to achieve complete cure in recurrent diseases [34].
The classical Sistrunk operation does not fully address the possible infrathyroid extension of the thyroglossal tract remnant that may occur in recurrent cases. Indeed, some authors have suggested that there is a similar branching pattern of accessory ducts that may be present in the infrathyroid region as typically found in the suprahyoid tissues. Consequently, some studies have recommended a deeper and further excision extending to the thyroid isthmus or the pyramidal lobe of the thyroid gland in recurrent cases of thyroglossal duct fistulas [35].

Patel et al., specifically suggested a wide local excision that includes a central 2-4cm of strap muscle down to the level of the pretracheal fascial plane. Following a similar concept, some authors have reported successful management of post-Sistrunk procedure recurrent thyroglossal duct remnants with en bloc central neck dissections [36].

As mentioned above a combined transoral/cervical approach was used successfully for a rare intralingual recurrence of a thyroglossal duct cyst [37]. Since these cases are not common, the authors did not recommend this approach to be a routine procedure but may be helpful for certain cases of thyroglossal duct remnants.

Perkins et al., also reported a combined transoral/cervical approach termed “suture-guided transhyoid pharyngotomy. This procedure was developed to address the challenges encountered when attempting complete excision of all epithelial tracts of the neck disease in recurrent TGDC cases. Basically a traction suture is placed transorally, which pulls the tongue base musculature into the cervical operative field. With the traction suture under tension, a core of tongue base tissue is excised in continuity with the foramen cecum mucosa. The advantage of this technique is reported to be the ability to perform precise and safe excision of suprahyoid and tongue base tissues with direct access [38].

A recent article by Maddalozza et al., described the concept of posterior hyoid space, which was demonstrated through cadaveric and patient dissections. The authors suggested that recurrent thyroglossal duct fistulas may occur as a consequence of incomplete resection in the peri-hyoid region and an understanding and removal of the central tissue in the posterior hyoid space may ensure a complete resection. Consequently, their surgical approach involves full exposure of the thyroid cartilage lamina and removal of tissue in the area posterior to the hyoid bone [39].

Only two studies discussed complications associated with managing recurrent thyroglossal duct remnants after failed Sistrunk procedures. Patel et al., mentioned that extended surgical dissection for recurrent cases may lead to higher complication rates due to the greater soft tissue dissection required but no specific complications or their rates were reported. Similarly, Perkins et al., stated that there were minor long-term issues with scarring and localized dermatitis in patients with recurrent thyroglossal duct fistula but no specifics or rates were reported [40,41].

In this study we have performed hyoid closure by approximating both cut ends of the hyoid bone to each other and fixation of them tightly in a vertical line of closure by 1 zero vicryl sutures 3 to 5 in number. So by this technique we close a substantial dead space created by removal of a portion of the hyoid bone and a core of tongue muscles. Again this space may harbor thyroglossal remnants which lose its connection to the subcutaneous space preventing recurrence but still possess its natural connection to the pharynx through the foramen cecum. Then we re approximate the strap muscles and suture them by vicryl sutures, followed by continuous stitch closure of the subcutaneous layer and skin closure by tight interrupted silk sutures or subcuticular Prolene stitch. So we obtain four layers of tight closure without leaving a drain after proper hemostasis.

So we avoided, an extended or wide local excision for the management of recurrent disease with its considerable risk of complications and still with unpredictable outcome.

Conclusion:

Management of recurrent thyroglossal duct remnants after a primary Sistrunk procedure or primary thyroglossal fistula remains a surgical challenge. Recurrences most often result from incomplete removal of thyroglossal duct remnants at the suprahyoid region. Other areas of recurrence include perihyoid, infrathyroid, and tongue base regions. Consequently, an extended or wide local excision for the management of recurrent disease may be necessary but with a considerable risk of complications and still with unpredictable outcome. So, tight hyoid gap closure followed by layered closure is a valuable option and effective surgical technique in the management of recurrent thyroglossal duct remnants and primary thyroglossal fistula for prevention of recurrence. Also we recommend the same technique for virgin cases with thyroglossal cyst to minimize or prevent recurrence.
References


