

Management Of Recurrent Thyroglossal Duct Remnants After Sistrunk Procedure: A Clinical Narrative Review Of Surgical Approaches

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Abstract

Background: Sistrunk operation for thyroglossal duct cysts has a high cure rate. There remains the rare patient, however, that will have recurrence despite a competently performed procedure. These patients present a surgical challenge since no standard treatment exists. **Objective:** To review and discuss the management options of recurrent thyroglossal duct remnants after a failed Sistrunk procedure. **Methods:** A comprehensive literature review of EMBASE, MEDLINE and Evidence Based Medicine Reviews databases was performed to identify the relevant articles. Following a thematic qualitative analysis, a clinical narrative review is presented. **Results:** Applied anatomy, embryology and histopathology proffered an extended or wide local surgical dissection to produce a definitive cures. Specific areas that required further excision included the suprahyoid, tongue base, infrahyoid, and posterior hyoid regions. **Summary:** Several different surgical methods have successfully managed recurrent thyroglossal duct remnants. Understanding the anatomy and pathophysiology is important in treating these challenging cases.

INTRODUCTION

Thyroglossal duct cysts (TGDC) are among the most common congenital neck masses noted in the pediatric population.¹⁻⁵ Subsequently, the removal of a TGDC is a commonly performed surgical procedure in children.⁶ The successful management of this midline neck mass is predicated upon a thorough understanding of the embryogenesis of the thyroid gland and the thyroglossal tract.⁷

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. 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 TGDCs.⁷

Historically, extremely high recurrence rates were reported with incision and drainage, and simple cyst excision procedures.⁸ The understanding and appreciation of the

embryology and pathophysiology of TGDCs 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, Schlang described a procedure that involved the excision of the mid-portion of the hyoid bone in continuity with the main neck mass.⁹ 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.^{10,11}

Sistrunk's principles of surgical management of TGDCs 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.^{4,12-15} Although resulting in a substantial reduction in the postoperative recurrence rates, many cases of TGDC continue to recur despite the practice of seemingly adequate surgical resection.¹⁶⁻¹⁸

For a TGDC 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.^{6,18,19} 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.

The objective of this review is to present the reported approaches used in the recurrent cases of TGDCs after a primary Sistrunk operation. Currently, there are several reports of managing these troublesome cases but no comprehensive review exists to date, which summarizes the differing approaches.

REVIEW

The databases of EMBASE, MEDLINE, and Evidence Based Medicine Reviews were searched using a comprehensive search strategy including the following terms: “recurrent”, “recurrence”, “thyroglossal duct”, “thyroglossal duct cyst”, and “thyroglossal duct tracts”. There were no time or language restrictions. The titles and abstracts were obtained for all studies identified by the search strategy and reviewed. Those studies that discussed management strategies for recurrent TGDCs after Sistrunk procedures were included for the review. The references of these studies obtained in full text for review were hand-searched for any additional relevant studies not identified by the original database search.

Due to the variability in treatments in the reviewed articles, along with the poor overall level of evidence, a formal systematic review or a meta-analysis was deemed not to be feasible. Instead a qualitative analysis using a common thematic coding was performed and a clinical narrative review is presented.

IMAGING

Only three studies were identified that explicitly discussed the role of imaging in those patients with recurrent disease after primary Sistrunk operation.²⁰⁻²² Two of the studies indicated that imaging techniques (ultrasound, magnetic resonance imaging, and fistulography) in these cases were not useful in analyzing the recurrent disease.^{21,22} That is, none of the tested modalities were helpful in identifying a remnant tract or cyst. Yet, in one case series of three patients, fistulography was found to be valuable in demonstrating the extent and anatomy of the remnant tracts.²⁰ More specifically, the residual suprahyoid tract up to the foramen cecum with side branches was clearly visualized. This allowed directed surgical resection, which led to definitive cures in all three patients.

Overall, there is no convincing evidence that imaging in those patients with post-Sistrunk recurrence is a useful measure at this time.

HISTOPATHOLOGY

The histopathological basis of thyroglossal duct is well described and many studies have reported on the varying features, which may account for the recurrent problems.⁷ Subsequently, surgical techniques have been designed to address these findings (see below).

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.^{1,6,16,18,23,24} More specifically, several accessory tracts and variable patterns of branching to the tongue base in the suprahyoid region have been reported.^{6,23} Some authors have described this arborization of the thyroglossal duct as a pattern resembling a “Christmas tree.”^{16,18} For this reason, it can be assumed that the TGDC in its entirety cannot be removed unless a wide core of tissue is excised along the path of the thyroglossal tract.

Traditionally, the wider resection approach was advocated by several authors in hopes of removing all of the branching remnants in cases of recurrent TGDCs.^{6,16,18,23} Hoffman et al. reported definitive cure in all recurrent patients with further, wider excisions.¹⁶ Their histological review showed high variability in suprahyoid patterns of drainage, with accessory tracts off 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 of TGDCs.

One study compared the number of accessory thyroglossal duct tracts in those patients who failed the Sistrunk operation versus those who did not recur.¹⁸ This study demonstrated that there were a greater number of accessory thyroglossal tracts in the former group, which again suggests that wider excision along the path up to the foramen cecum is recommended in recurrent disease.

SURGICAL TECHNIQUES

Historically several authors advocated for the Sistrunk operation for recurrences after aspiration, incision and

drainage, and simple cyst excision procedures.^{6,17} Over time, few articles have addressed the management approaches after a failed Sistrunk procedure.^{6,8,16,18,19,25,26}

HYOID BONE AND SUPRAHYOID RESECTION

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 report removing 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.^{6,18,25} Similarly, Hoffman also suggested that a wider excision of the suprahyoid tissue up to the level of the pharyngeal mucosa with a margin of tongue base muscle be included in recurrent cases.¹⁶ As well, he recommended that skeletonizing the tract above the hyoid bone should be condemned, since it does not account for the accessory tracts.

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.⁸ Subsequently, the revision operation included additional hyoid bone removal and suprahyoid core of tissue excision to the foramen cecum and good cure rates were demonstrated afterwards. Moreover, this study recommended that a 0.5 cm radius core of tissue from midportion of the hyoid bone to tongue base be removed.

In practice, excision of the suprahyoid tissue can be quite challenging, since the tract is ill defined^{24,27} and some have proposed that this area may actually represent a pseudocyst rather than a true thyroglossal duct remnants.^{23,28}

There is some controversy regarding the amount of hyoid bone that requires excision to prevent recurrences. Most authors report that the central 2/3rd of the hyoid bone or the area between the inferior cornuae must be removed to reduce the risk of leaving tracts in situ but this has not been proven.^{18,25}

FORAMEN CECUM AND TONGUE BASE MUCOSA

Some authors have advocated the excision of the foramen cecum itself to prevent further recurrences^{22,27}, while others

suggest that entry through the tongue base into the oropharynx is not necessary for definitive cure.^{6,18,25} One study reported a combined transoral/cervical approach to remove a core of tissue around the foramen cecum for a rare intralingual cyst recurrence.²² 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.

INFRAHYOID RESECTION

The classical Sistrunk operation does not fully address the possible infrahyoid 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 infrahyoid region as typically found in the suprahyoid tissues.^{7,19,25} 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 TGDCs.^{7,18,25,29} Patel et al. specifically suggested a wide local excision that includes a central 2-4 cm of strap muscle down to the level of the pretracheal fascial plane.²⁵

Following a similar concept, some authors have reported successful management of post-Sistrunk procedure recurrent TGDCs with en bloc central neck dissections.^{19,26}

NOVEL TECHNIQUES

As mentioned above a combined transoral/cervical approach was used successfully for a rare intralingual recurrence of a TGDC.²² 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.”²⁶ 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.²⁶

A recent article by Maddalozza et al. described the concept of posterior hyoid space, which was demonstrated through cadaveric and patient dissections.³⁰ The authors suggested that recurrent TGDCs 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.

COMPLICATIONS

Only two studies discussed complications associated with managing recurrent TGDCs 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.²⁵ Similarly, Perkins et al. stated that there were minor long-term issues with scarring and localized dermatitis in patients with recurrent TGDCs but no specifics or rates were reported.²⁶

SUMMARY

Management of recurrent TGDCs after a primary Sistrunk procedure 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, infrahyoid, and tongue base regions. Consequently, an extended or wide local excision is recommended for management of recurrent disease (Table 1).

Figure 1

Table 1. Summary of the surgical methods used to manage recurrent thyroglossal duct remnants after failed Sistrunk operation.

<ul style="list-style-type: none">• Extended local excision of central suprahyoid soft-tissue up to base of tongue, including musculature of tongue base (+/- foramen cecum and tongue base mucosa)• Additional removal of hyoid bone at lateral cut-ends (remove at least 2/3rd of the midportion)• Extended local excision of infrahyoid soft-tissue down to the level of thyroid isthmus or pyramidal lobe of thyroid gland (if present)• Wide local excision of soft-tissue from the space posterior to the hyoid bone
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