Is the Treatment for Thyroglossal Duct Cysts Too Extensive?

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During the fourth week of fetal development, the thyroid gland develops from a median endodermal thickening in the floor of the primitive pharynx caudal to the future tuberculum impar. This thickening descends in the neck as a bilobular diverticulum between the first and second branchial arches. This point corresponds to the foramen cecum in adult life. The hyoid bone is simultaneously formed from the second and third branchial arches growing from behind forward and dividing the tract into infrahyoid and suprahyoid portions. By the end of the seventh week, the thyroid gland has normally reached its final location anterior to the trachea. Usually, any connection between the cervical thyroid and its point of origin at the foramen cecum is obliterated and disappears. The original opening of the duct, however, remains as a vestigial pit, the foramen cecum. Should the duct or a remnant persist, cyst formation may occur anywhere from the base of the tongue to the thyroid isthmus [7].

The thyroglossal duct cyst is a rare but occasional cause of a benign midline neck mass. It most often occurs in the pediatric age group, but may be found in the older population as well. Thyroglossal duct cysts most often present with a palpable asymptomatic midline neck mass at or below the level of the hyoid bone. The spectrum of clinical symptoms may be as varied as the multiple forms of surgical treatment.

Since Sistrunk’s [2] original treatise in 1920 on the pathogenesis and management of thyroglossal duct cysts, minimal attention has been directed to the long-term results of this procedure or the validity of thyroglossal duct cyst embryogenesis. Controversy continues among surgeons concerning the management of this entity. Some believe the Sistrunk procedure is too radical and that simple excision of the cyst is sufficient treatment. Patients subjected to this latter form of therapy have reported recurrence rates that range from 50 to 68 percent [3,4]. Others advocate excision of the cyst and the infrahyoid tract, a technique that has also met with an unacceptably high failure rate. In his classic publication, Sistrunk [2] reported his experience with 31 patients with thyroglossal duct cysts and described the technique of excision of the cyst, infrahyoid tract, body of the hyoid bone, and a core of tissue one eighth of an inch on all sides of the suprahyoid portion of the tract to the foramen cecum.

The purpose of this study is to report our long-term results with thyroglossal duct cysts and determine if the classic Sistrunk operation is a valid form of surgical treatment or represents surgical overkill.

Material and Methods

Between 1970 and 1985, 64 patients were treated for thyroglossal duct cysts at the University of Oklahoma Health Sciences Center. Their ages ranged from 1 to 63 years, with a mean of 12.4 years. The male to female ratio was 43:21 and 46:19, respectively. The duration of symptoms before operation ranged from 1 day to 12 years, with a mean of 10.1 months. Information for this study was obtained from hospital records, telephone interviews with patients or family members, and questionnaires. Statistical analysis was performed using the chi-square test with a significance level of p < 0.05. Postoperative follow-up ranged from 1 month to 15 years (mean 4.8 years). All diagnoses were confirmed by the presence of a midline cystic cervical mass, the microscopic presence of thyroid epithelium or follicles in the cyst wall, or both.

Patients were divided into four groups for the study. Group A comprised 37 patients (58 percent) who underwent the classic Sistrunk operation (resection of the cyst, tract, and midportion of the hyoid bone, and tract excision to the foramen cecum). Group B comprised 20 patients (31 percent) who underwent a modified Sistrunk operation (resection of the cyst, infrahyoid tract, and midportion of the hyoid bone). Group C comprised patients who underwent excision of the cyst and infrahyoid tract. There were no patients in this group. Group D comprised seven patients who received excision of the cyst only (11 percent).

Results

The majority of these patients (70 percent) were initially asymptomatic and presented for evaluation of a midline neck mass discovered incidentally by the patient or a family member. The most common presenting complaints were pain (16 percent), a draining sinus tract (16 percent), infection (5 percent), and dysphagia (3 percent). The length of the hospital stay varied depending on the patient’s symptoms. A thyroidectomy was performed on patients with complications (e.g., infection or drainage). Fifty-eight percent of patients underwent drainage of the cyst.

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Treatment of Thyroglossal Duct Cysts

A thyroid scan was performed on 25 patients (39 percent) preoperatively, and all of the findings were normal with no evidence of ectopic thyroid tissue and the gland at the normal location. Fifty-eight patients (91 percent) had not undergone previous operation for the treatment of thyroglossal duct cysts. Of the six patients (9 percent) with a history of operation, four (6 percent) underwent excision of the cyst only and two (3 percent) had incision and drainage of the cyst.

Forty-nine patients (77 percent) had cysts presenting in the midline, 11 (17 percent) to the left of the midline, and 4 (6 percent) to the right of the midline. Twenty-nine patients (45 percent) were found to have cysts at the level of the hyoid bone. Of the remaining 34 patients, 9 (14 percent) had cysts located above the hyoid bone and 25 (39 percent), below it. Eleven patients (17 percent) were found to have evidence of infection at the time of operation. Twenty-eight patients (44 percent) received antibiotics in the perioperative period. Eleven patients (17 percent) had rupture of the cyst intraoperatively. Drains were placed in 46 patients (72 percent) and left in place for 24 to 48 hours. In all, the midportion of the hyoid bone was resected in 56 patients (88 percent).

In the excised specimen, three basic types of epithelium were identified: squamous, cuboidal, and columnar. In 10 patients, no epithelial lining was found. This latter finding has been previously observed in those patients with long-standing inflammation [5]. One 51 year old patient with an infrahyoid thyroglossal duct cyst located to the left of the midline contained a mixed papillary and follicular carcinoma. He was returned to the operating room 2 weeks later where he underwent a left thyroid lobectomy and isthmusectomy.

Postoperative complications, occurring in 13 of 64 patients (20 percent), included wound infections in five patients, pulmonary problems in three patients, wound seromas in two patients, and a wound hemato ma, thrombophlebitis, and gastroenteritis in one patient each. Age did appear to be a factor in the development of postoperative complications. Patients less than 10 years of age or greater than 60 years had a higher number of postoperative complications compared with the middle-aged group. The incidence of wound infections observed in children less than 10 years of age (14 percent) was significantly higher (p < 0.05) than the incidences in the older age groups. Patients with active infection or draining sinus tracts had a higher complication rate (27 percent) compared to patients without infection or drainage (19 percent) (p not statistically significant).

Fifty-eight patients (91 percent) were available for long-term follow-up. The study period ranged from 1 month to 15 years with a mean of 4.8 years. Of the patients available for long-term follow-up, 36 were in Group A, 16 were in Group B, and 6 were in Group C. The follow-up period was further broken down into early and late periods, with the early period being the first 6 months after operation. Using these time periods, there were three recurrences (5 percent) at 2 months, 3 months, and 4 months. Of these patients, two were in Group B (cyst excision only) and one was in Group A (classic Sistrunk operation). Thus, Group A patients were found to have a 2.7 percent incidence of recurrence in the early follow-up period compared with a 33 percent incidence of recurrence in Group D (statistically significant, p < 0.05). Only one of these recurrences was in the group less than 10 years of age, with the two remaining recurrences in the middle-aged group. All early recurrences took place in patients whose cyst had been located in the infrahyoid position. Rupture of the cyst at the time of the original procedure was associated with an 11 percent incidence of recurrence compared with a 4 percent incidence among patients whose cyst had not been ruptured (not statistically significant). Recurrence rates among patients with infection or draining sinus tracts (10 percent) were found to be slightly higher than those of patients without infection (4 percent).

Comments

A thyroglossal duct cyst, a midline neck mass, can occur in any age group but is more common in the younger age group, and males are more often affected. The differential diagnosis of a midline neck mass includes a pyramidal lobe of the thyroid, thyroid adenoma, aberrant thyroid tissue, a branchial cleft cyst, lipoma, dermoid cyst, lymphadenopathy, and hemangiomas [6, 7]. In this study, 67 percent of the patients were male, and the mean age of the patients treated (12.4 years) was similar to those previously reported [2, 6-10]. The cyst most often presented as an asymptomatic midline neck mass at or below the hyoid bone which moved with deglutition [7, 9-11]. When a thyroglossal duct cyst reaches a large size, it may cause symptoms of dysphagia or mild respiratory distress. Up to 50 percent of thyroglossal duct cysts will eventually become infected [6]. Once infected, they will frequently drain spontaneously or require incision and drainage. Of those patients who undergo a drainage procedure, the recurrence rate may be as high as 55 percent [4]. In our patient population, two patients had previously undergone incision and drainage of an infected thyroglossal duct cyst.

Whether all patients scheduled to undergo operation for a thyroglossal duct cyst should have a preoperative thyroid scan to rule out an ectopic thyroid gland continues to be debated [6, 7, 10, 12]. This situation usually manifests itself as the child grows and the small aberrant gland is unable to keep pace with the increasing metabolic demands of the patient.
The continued stimulation of thyroid-stimulating hormone causes the gland to hypertrophy and become symptomatic. Should the diagnosis be made before operation, the patient should receive thyroid hormone replacement therapy, which should cause regression of the ectopic gland by suppressing thyroid-stimulating hormone [12]. If the diagnosis is made at the time of operation, the gland should be divided in the midsagittal plane and implanted under the strap muscles [13]. Should the excision of an ectopic thyroid gland occur and not be recognized, the patient may not manifest symptoms of hypothyroidism for some time. Detailed follow-up is mandatory to ensure adequate thyroid hormone replacement.

In 1893, Schlange [14] proposed excision of the central portion of the hyoid bone in the management of thyroglossal duct cysts. This simple maneuver reduced the recurrence rate to 20 percent [4]. In 1920, Sistrunk [2] reported his experience with 31 patients with a thyroglossal duct cyst. The recurrence rate in his series was only 3 percent. Sistrunk stressed two basic principles in his initial monograph that he believed were essential in the management of thyroglossal duct cysts. One principle was that the thyroglossal duct usually passes through the hyoid bone although it may pass above or below it. In order to ensure excision of the duct, Sistrunk advocated excision of the hyoid bone. This principle remained unchallenged until 1977, when Ellis and Van Nostrand [15] examined 200 serially sectioned adult larynges and 30 embryos at various stages of development. In that classic study, they determined that the tract bears a constant anterior relationship to the hyoid bone and hooks around the sharp inferior border of the bone to lay in the concavity on the posterior surface before descending in the neck as a result of early remodeling of the bone. The investigators concluded that the removal of the midportion of the hyoid bone was necessary since the tract and bone are intimately related. Sistrunk’s second principle was that no attempt should be made to isolate the tract above the hyoid bone, since it is usually small and friable. Thus, any attempt to locate the duct would be wrong since, if broken, it would be difficult if not impossible to remove. To avoid this complication, Sistrunk advocated removal of the duct and the surrounding tissues for a distance of one eighth of an inch on all sides of the duct from the hyoid bone to the foramen cecum [2]. Other investigators validated this point by histologically examining the tissues above the hyoid in patients with a thyroglossal duct cyst. Soucy and Penning [5] reviewed 44 cases of thyroglossal duct cyst and found that the suprahoid tract was often multiple, could arborize frequently and could even be discontinuous. Of the patients examined, 24 (55 percent) were found to have multiple ducts. In many instances, the surgeon may believe the duct is visible when no tract can be identified histologically.

In recent years, researchers have attempted to identify those factors associated with increased rates of recurrence after excision of a thyroglossal duct cyst. Hawkins et al [7] studied 53 patients who underwent surgical excision of thyroglossal duct cysts and concluded that young patient age, skin involvement, lobulation of the cyst, rupture of the cyst at operation, and failure to follow the second principle of Sistrunk were factors that contributed to an increased recurrence rate. Ein et al [4], in their review of 270 patients who underwent surgical treatment for thyroglossal duct cysts, found that infection and draining sinus tracts were the most important factors in determining recurrence.

In the present study, there were three early recurrences. The first patient was a 23 year old woman who had cyst excision only. The procedure was uncomplicated. The second patient was a 12 year old boy who had cyst excision only, but rupture of the cyst occurred during operation. The third patient was a 4 year old boy with a recently infected cyst who underwent a standard Sistrunk operation. Thus, all patients had risk factors, as described by Sistrunk, that contributed to recurrence, those being rupture of the cyst, infection, and young patient age.

Malignant transformation of thyroglossal duct cysts is a rare but well described pathologic entity. The first case of thyroglossal duct carcinoma was reported in 1915 by Uchermann [16]. There have only been 100 cases reported in the literature [7,17]. The diagnosis is seldom made preoperatively (as was true in this study) since the foci are usually small, and in most cases, the thyroid function test and scan results are normal. The majority of cases are found in women, and between 75 to 89 percent are papillary carcinoma. Other cell types, such as follicular carcinoma and mixed papillary and follicular carcinoma, are much less common [17,18]. Magsalin et al [17] suggested two theories of origin of the carcinoma when it is found in the thyroglossal duct. It either represents spreading of an occult thyroid carcinoma or the primary carcinoma arises in ectopic thyroid tissue.

The correct form of treatment for thyroglossal duct carcinoma is ill defined because of limited experience. Most surgeons concur that the Sistrunk procedure is adequate treatment for the disease [17], and others take a more aggressive approach, including total thyroid exploration with multiple biopsies to near-total thyroidectomy, radioactive iodine ablation, and thyroid suppression [17].

Summary

From 1970 to 1985, 64 patients underwent surgical management for thyroglossal duct cysts at the University of Oklahoma Health Sciences Center. Their ages ranged from 1 to 63 years with a mean of 12.4 years. Ninety-one percent of the patients were available for follow-up. All patients underwent one of three operations. The majority place him in the midportion of the hyoid bone. The periparotid area was site of the injection. In the majority of cases, the tract was successful in the neck, leaving the thyroid gland intact.
of three forms of treatment: the classic Sistrunk operation, a modified Sistrunk operation, or excision of the cyst only. Eleven percent had undergone a previous procedure. The majority of patients were found to have a cyst in the midline at the level of the hyoid bone. Eighty-eight percent of the patients underwent excision of the midportion of the hyoid bone. There were no postoperative deaths and the perioperative complication rate was 20 percent, the majority being wound-related. All recurrences took place within 4 months of operation. Factors that appeared to be associated with an increased risk of complications and recurrence included young patient age (less than 10 years), rupture of the cyst at the time of operation, infection, and failure to excise the midportion of the hyoid bone and the suprahyoid tract. One patient was found to have a mixed papillary and follicular carcinoma in the thyroglossal duct. We support the original premise of Sistrunk that “the cure of thyroglossal cyst is unsuccessful unless the epithelium-lined tract, running from the cyst to the foramen cecum is completely removed,” including the hyoid bone [2].

References

Discussion
Jon S. Thompson (Omaha, NE): Dr. Bennett, your study very nicely points out the importance of technical details in the outcome of surgical procedures. I agree with your conclusion because my training and experience have led me to believe that the Sistrunk operation is the best procedure for this condition, but looking at the data objectively, can you really make that conclusion in your study? Certainly, you can make the statement that simple cyst excision is inadequate treatment based on the very high recurrence rate. Also, the conclusion can be made that removing the hyoid bone is important for preventing recurrences. But as I look over the data, there were no recurrences with the modified Sistrunk operation. Thus, we have shown that excision of the suprahyoid tract up to the foramen of the cecum is really an important factor based on these data. However, other studies have shown this, as you mentioned.

You have identified a number of other factors that seem to be important in recurrence rates, such as the age of the patient, cyst rupture, and infection. Were you able to compare your different treatment groups to see if, in fact, they were comparable in regard to the other factors that may affect recurrence? Finally, Dr. Bennett, what is your strategy for midline masses? Do you aspirate them? What do you do if you remove the thyroid inadvertently?

Keith G. Bennett (closing): Dr. Thompson, there was one recurrence in the group of patients who underwent the Sistrunk operation and no recurrences in the 20 patients who had the modified operation. The small sample size of this latter group would, in my opinion, limit our ability to interpret the data. I suspect that if there had been more patients in the modified Sistrunk group, there would have been some recurrences.

On occasion, while exploring the neck for a presumed thyroglossal duct cyst, one may instead find ectopic thyroid tissue; however, ectopic thyroid tissue is not usually recognized preoperatively but is instead found incidentally during operation. The tissue will typically be deep purple like normal thyroid tissue. There are two treatment options. The gland can either be divided along the mid sagittal plane and implanted underneath the strap muscles laterally, making every attempt to preserve the blood supply, or since the current treatment for hypothyroidism is both safe and effective, it can simply be excised and the patient can be given full thyroid hormone replacement therapy. The results are usually excellent with either treatment option.