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The Effect of Complete Excision of the Hyoid Bone Corpus from the Cartilage Margins on the Success of Sistrunk Procedure in Children: The Preliminary Results

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ABSTRACT

Objective: To analyze the effects of the hyoid bone excision magnitude and Z-shaped extra stitches, putting in the base of the mouth on the success of the surgical treatment of thyroglossal duct cyst (TGDC).

Materials and Methods: A retrospective analysis of 19 patients who underwent Sistrunk procedure between October 2013 and November 2018 was performed, and 16 patients with sufficient data were included in this study. In surgical procedures, we modified the Sistrunk procedure with a broader excision of mid-portion of hyoid bone, and Z-shaped extra stitches were put in the floor of the mouth. The patients were evaluated for age, sex, preoperative examinations, surgical findings, complications, and recurrence.

Results: The mean age of the patients was 5.12 years (range, 2 to 9 years), and 11 (69%) were female, and five (31%) were male. Twelve (75%) and three (19%) patients were presented with cyst and fistula formation, respectively. One (6%) patient referred to us as a result of recurrence with her previous sonographic results, which indicated TGDC. Only one patient (6%) experienced disease recurrence, and it was achieved a success rate of 94%. All the patients were followed for a mean of 12.81 (range, 6 to 21 months) months.

Conclusion: Extensive removal of the hyoid bone and Z-shaped sutures ligating possible ducts associated with the oral cavity may increase the rate of success in the surgical treatment of TGDC in children. However, this suggestion should be supported by further investigations.

Keywords: Thyroglossal duct cyst, Sistrunk operation, hyoid cartilage, recurrence

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INTRODUCTION

Thyroglossal duct cyst (TGDC) is the most common developmental neck cyst in childhood (1) and accounts for approximately 75% of all surgical midline neck masses (2). TGDC is usually defined as an anterior midline neck mass, which is mobile and painless (3). The simultaneous movement of the cyst with the hyoid bone while swallowing is generally considered a reliable diagnostic marker (4). The removal of the middle one-third of the hyoid bone with cyst (Sistrunk procedure) is offered as the gold standard surgery (5, 6). Although it has been a procedure using for nearly a century, it has been reporting a recurrence rate ranging from 1.2% to 20% (3, 7). The Sistrunk procedure has been modified in several ways to reduce recurrence rates and make surgery even easier (8, 9).

In this study, we analyzed the effects of the hyoid bone excision size and Z-shaped extra stitches, putting in the base of the mouth on the success of the surgical treatment of thyroglossal duct cyst.

MATERIALS and METHODS

The data of 19 patients who underwent Sistrunk surgery for TGDC in Sütçü İmam University Health Practice and Research Hospital between October 2013 and November 2018 were studied retrospectively after having been obtained the approval of the local ethics committee which is adherence to the Declaration of Helsinki. Two patients who were lost in the postoperative follow-up and one girl with missing data were excluded from this study. In total, 16 patients included in this study. The patients were evaluated for age, sex, preoperative examinations, surgical findings, complications, and recurrence.

The corpus of hyoid bone was excised with monopolar cautery from the margins of cartilage after the muscles were released (Fig. 1). Then, the duct was ligated without excising the lingual mucosa, and the specimen was achieved (Fig. 2). Finally, while the index finger of the surgeon was in the foramen cecum, lingual mucosa was sutured by two superficial Z-shaped stitches considering possible accessory ducts (Fig. 3).

Statistical analysis was performed using SPSS 17.0 (IBM Corporation, Armonk, New York, USA). Quantitative data are expressed as mean and range (min, max).

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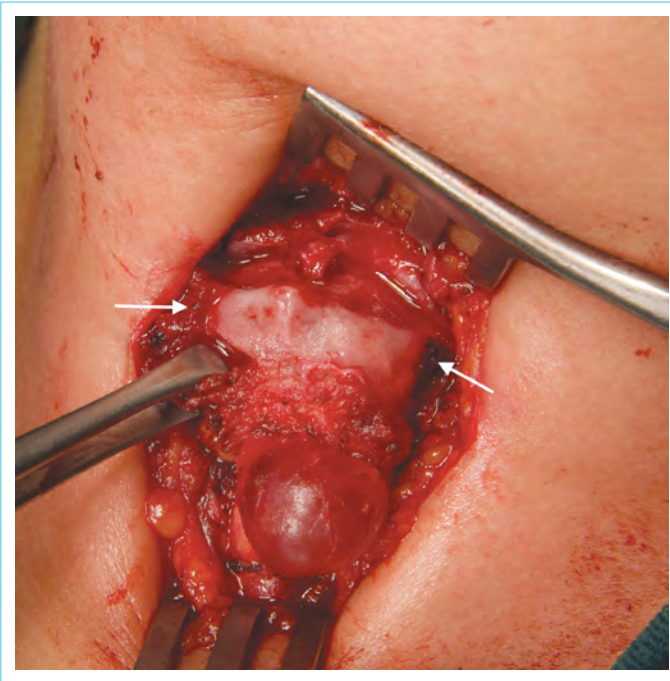


Figure 1. Margins of the epiphyseal cartilage (arrows)

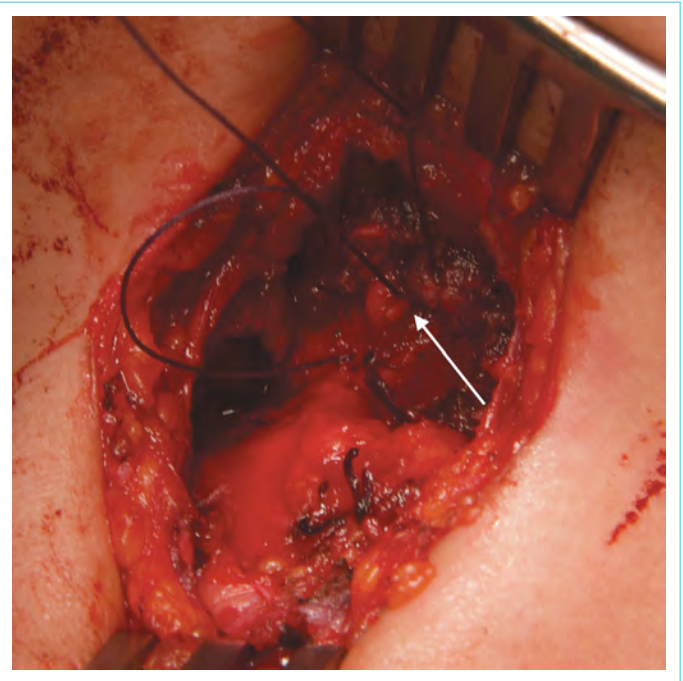


Figure 3. Z-shaped extra stitches (arrow)



Figure 2. Excised specimen

RESULTS

Of the 16 patients with a mean age of 5.12 years (range, 2 to 9 years), 11 (69%) were female, and five (31%) were male. Twelve (75%) and three (19%) patients were presented with cyst and fistula formation, respectively. One (6%) patient referred to us as a result of recurrence with her previous sonographic results, which indicated TGDC. The sonographic investigation has confirmed the diagnosis of TGDC in 12 patients, and no diagnostic modality was considered necessary in three patients with fistula formation.

Only one patient (6%) described previous cervical infection requiring antibiotherapy before surgery. We performed a successful Sistrunk procedure in 15 patients. All patients were discharged on the first postoperative day. The patient (6%) who presented with recurrence underwent cystectomy, hyoid bone excision from the margins of epiphyseal cartilage and resection of the main duct with the surrounding tissue. After three weeks of our surgery, a boy (6%) experienced recurrence. Finally, the success rate of 94% was achieved. The recurrent case was re-operated at 12 months postoperatively. It was detected incomplete hyoid bone resection and convergence (not union) in two parts of the resected hyoid bone. All the patients were followed for a mean of 12.81 (range, 6 to 21 months) months. Excellent cosmetic results were observed. The characteristics of the patients are summarized in Table 1.

DISCUSSION

The main surgical principle of TGDC is resection of the cyst and the center of the hyoid bone. Sistrunk, in his last publication, recommended resection of cyst with a mid-portion of the bone nearly one-fourth of an inch in length while foramen cecum and lingual mucosa were intact. Various publications have been reported, modifying the Sistrunk procedure, on the size of the hyoid bone to be removed (3, 9, 10). All these efforts aim to increase the success rate without a recurrence. However, recurrence rates up to 20% are reported in the literature (3, 7). One of these reasons is attributed to that the proximal duct extending from the mid-portion of the hyoid bone to the foramen cecum is not visible and palpable (11, 12). The other reason is accessory ducts behind the corpus of the hyoid bone, which was well-described years ago by Horisawa et al. (13), associated with many secretory glands and oral mucosa. Therefore, we assumed that the extensive removal of the center of the hyoid bone (between both epiphyseal cartilage margins of the hyoid bone) and resection of the duct approximately one centime-

Table 1. The patients' characteristics and results

No	Gender	Age (years)	Type	Previous infection	Complication	Follow-up (month)
1	M	3	Cyst	-	-	17
2	F	5	Cyst	-	-	21
3	F	6	Cyst	-	-	13
4	F	3	Cyst	-	-	9
5	M	6	Cyst	-	-	10
6	F	2	Recurrence	-	-	9
7	F	3	Cyst	-	-	17
8	M	5	Fistula	-	-	12
9	F	9	Fistula	+	-	12
10	M	5	Cyst	-	Recurrence	14
11	F	6	Cyst	-	-	19
12	F	5	Cyst	-	-	14
13	M	7	Cyst	-	-	13
14	F	8	Fistula	-	-	8
15	F	5	Cyst	-	-	6
16	F	4	Cyst	-	-	11

F: Female; M: Male

ter in diameter with surrounding tissue would reduce the possibility of leaving these accessory and main ducts in place. Moreover, we stitched up the base of the mouth with two superficial Z-shaped absorbable sutures that could ligate possible undetected accessory branches.

With the described way of TGDC, we had a success rate of 94%. In the follow-up period, we experienced recurrence only in one patient (6%). Although this recurrence rate is within the range of the reported literature data (1.2–20%), the increasing number of patients will increase the validity of the result. Recurrence occurs in the first postoperative year in most patients (14). However, it was suspected that a technical problem in surgery might be responsible for our early recurrence. The second operation was performed one year after the first operation at the request of parents concerned about an additional procedure although we recommended an earlier intervention. In the second surgery of this child, we noticed incomplete resection of the hyoid bone in the previous surgery, and convergence (not union) in two separate parts of the resected hyoid bone. We observed the same findings in the secondary procedures of the other patient who presented with recurrence. The secondary cyst was removed, and hyoid bone resection was extended, as we described in both patients. Then the tissue between the center of hyoid bone and foramen cecum (including duct) was resected, and recurrence was not observed in the follow-up.

Ryu et al. (9) described and classified the resection margin of hyoid bone with computed tomography (CT) before surgery. In their classification, the terms of non-fusion, partial fusion, and complete fusion were used to express the state of epiphyseal cartilage between the corpus and greater horn of the hyoid bone. They reported no complete fusion in any of their pediatric patients. Thus, they

could divide the corpus of hyoid bone from greater horns (cartilage division technique) using monopolar cautery, not using a bone cutter. Consistent with their findings, we noticed this cartilage margin during the extensive removal of the mid-portion of the hyoid bone and were able to easily divide the hyoid corpus from greater horns using monopolar cautery. The presence of epiphyseal cartilage in children facilitates hyoid bone excision, as well as determines the excision boundaries at the hyoid bone.

Ultrasonography (US) is usually the first preoperative investigation in the presence of clinical suspicion of TGDC. CT and magnetic resonance imaging (MRI) make a clear visualization of the cyst and private anatomy of the patient's neck possible (15). However, radiation and anesthesia exposure during CT and MRI, respectively, are the main concerns in children. The US is the only imaging modality using in the present study, and pathologic examinations have confirmed the diagnosis of TGDC in all patients. If the diagnosis cannot be clarified with USG, CT, and MRI can be used to exclude dermoid cyst, branchial cyst, cystic hygroma, and thyroid masses (3). However, we believe that no additional diagnostic investigation other than the US is usually needed in the pediatric population. Before the secondary surgeries, we did not consider any additional imaging modality necessary as the pathological specimens in the first surgeries were confirmed TGDC, including thyroid tissue remnants. However, it should be kept in mind that MRI may help to specify a residual tract in recurrent disease (15).

The main limitations of this study are its retrospective nature and the inadequate number of patients. A prospective study with a sufficient number of patients may demonstrate the effectiveness of the technique.

CONCLUSION

We believe that preoperative physical examination and ultrasonography are sufficient for the diagnosis of TGDC in most of the cases. Sistrunk procedure is still the gold standard in the surgical treatment. Extensive removal of the hyoid bone with the duct approximately one centimeter in diameter with surrounding tissue and Z-shaped sutures ligating possible ducts associated with the oral cavity may increase the rate of success in the surgical treatment of TGDC in children. However, this suggestion should be supported by further investigations.

Ethics Committee Approval: The Sütçü İmam University Faculty of Medicine Clinical Research Ethics Committee granted approval for this study (date: 03.07.2019, number: 14).

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – AEK, ABD, AGG; Design – ABD, AEK; Supervision – AEK, AGG; Resource – AEK, AGG; Materials – AEK, ABD, AGG; Data Collection and/or Processing – AEK, ABD, AGG; Analysis and/or Interpretation – ABD; Literature Search – ABD, AGG; Writing – ABD; Critical Reviews – AEK, ABD, AGG.

Conflict of Interest: The authors have no conflict of interest to declare.

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