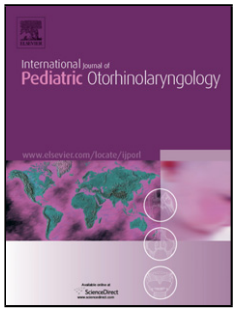

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Modified Sistrunk Operation: New Concept for Management of Thyroglossal Duct Cyst

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Running Title: A hyoid cartilage division method for TGDC

Abstract

Objective: To describe a method of hyoid cartilage division during Sistrunk operation for management of thyroglossal duct cyst (TGDC) and compare postoperative outcomes with those of conventional hyoid bone cutting.

Methods: Fifty-nine patients who received operative treatment for TGDC from January 2005 to July 2013 were enrolled. The degree of fusion in the cartilaginous portion of the hyoid was evaluated by preoperative neck computed tomography and classified into (1) non-fusion, (2) partial fusion, and (3) complete fusion. Techniques of hyoid management, division of the cartilaginous portion of the hyoid or conventional hyoid bone cutting, during Sistrunk operation were analyzed according to the fusion classification.

Results: Of the 59 patients, 27 were pediatric patients and 32 were adults. In 32 adults undergoing TGDC, there were 13 cases (41%) of complete hyoid fusion and 7 (22%) of non-fusion. A total of 20adult patients (63%) were treated with the cartilage division. Among the pediatric patients, 96% of cases were classified as non-fusion of the hyoid, and all of these were treated with the cartilage division. Patients who were treated with the cartilage division showed better postoperative results. However, there were no statistical differences.

Conclusions: Our cartilage division technique at Sistrunk operation for TGDC is feasible and comparable to conventional hyoid cutting with regard to postoperative results.

Key Words: Thyroglossal duct cyst, Sistrunk operation, hyoid, ossification, bleeding

Introduction

Thyroglossal duct cyst (TGDC) is one of the most common congenital neck lesions, accounting for about 70% of midline neck masses in children.[1, 2] The reported incidence of TGDC or its remnant tract is approximately 7% in the general population,[3, 4] and a 15–17% rate of TGDC has been observed in published autopsy studies.[4, 5] TGDC is usually diagnosed before adolescence;[1, 6] however, it can be recognized in all age groups.[7] Therefore, even low-volume ENT surgeons may commonly encounter TGDC in any clinical setting.

Although advanced operating techniques for TGDC excision, including endoscopic or robotic management, have been introduced, and related articles have been published,[8-11] the classical "core principle" remains invariable. According to this principle, in TGDC operations, the central portion of the hyoid bone and the remnant dissection with core tissue should be resected with the cystic portion to prevent recurrence of TGDC.[12] Because the high-end advanced procedures have been accessible exclusively to a limited number of head and neck surgeons in specialized tertiary centers, in regular practice, the conventional Sistrunk operation or a modification thereof, is usually selected and performed for the treatment of TGDC.

During the conventional Sistrunk operation, the mid-portion of the hyoid body is cut off at the medial side of the lesser horn of the hyoid, usually with bone cutting forceps.[13] However, in cases without mature ossification of the hyoid bone,[14, 15] a non-fused cartilage portion between the body and the horn of hyoid can be easily dissected and divided by monopolar Bovie electro-cauterization or a scissors. These non-fused cartilaginous portions of hyoid bone are easily identifiable on preoperative neck computed tomography (CT).Therefore, as per the findings of preoperative neck CT scans; we have conducted cartilage division for removal of the central hyoid bone during Sistrunk operation in pediatric TGDC cases. Herein, we introduce a simple technique of hyoid cartilage division for the management of the body of the hyoid during Sistrunk operation and we evaluate its feasibility in the treatment of patients with TGDC.

Patients and Methods

Patients

Medical records of patients who underwent surgical treatment by a single surgeon (J.H.H) for anterior neck mass confirmed pathologically as TGDC at Seoul National University Hospital from January 2005 to July 2013 were retrospectively reviewed for the current study. This study was approved by the Institutional Review Board of Seoul National University Hospital (approval number: 1401-068-550).

Patients who were referred from other hospitals for treatment of recurrent or remnant TGDC after primary surgery, those in whom the body of the hyoid bone was not resected during TGDC excision, those who underwent a transoral approach or endoscopic approach to TGDC excision, those treated with a combined other head and neck surgery, and those with unavailable preoperative CT scans were excluded from the study. Thus, among 67 patients who received surgical treatment for TGDC during the study period, 2 revisions, 1 cases of hyoid preservation, and 5 cases without preoperative CT scan were excluded. Finally, 59 patients with TGDC were enrolled in the study.

Operative technique

The procedure of hyoid cartilage division is a simple modification of the conventional Sistrunk operation (Figure 1).[13]

1. A transverse incision, approximately 4 cm in length, was made in a skin crease inferior to the lesion and the skin and platysma muscle were reflected. Superiorly, a myocutaneous flap was elevated adequately to expose the hyoid bone and the cyst lying within connective tissues of the geniohyoid muscles. Inferiorly, the flap was extended to the level of the thyroid notch.
2. The strap muscles were divided at the midline or raphe and the cyst was dissected from surrounding tissues up to the hyoid bone. Then, under traction of the central hyoid with Allis grasping forceps, the attached muscles of the hyoid were dissected from the center to the lateral hyoid using monopolar Bovie electro-cauterization. During this step, a 1 cm width of geniohyoid muscle at the center of hyoid should be kept undissected until the next step, because the tract between the TGDC at the hyoid and the foramen cecum can be cut and lost during the dissection.
3. In patients with non-fusion of the hyoid, the cartilaginous junction between the hyoid body and the greater horn (the lesser horn is usually fused with the greater horn) was formed with a synovial joint and easily detected

during dissection of the attached muscles of the hyoid. With additional anterior traction of the hyoid at one side, the cartilaginous end of the body of the hyoid became more prominent and the non-fused cartilaginous portion was easily divided with dissection of surrounding dense fibrous strips by monopolar cauterization. The same procedure of hyoid cartilage division was then performed at the other side of the non-fused cartilaginous portion of the hyoid (Figure 2).

4. After complete cartilage division on both sides of the hyoid, the body of hyoid with tract or the cystic portion of the TGDC and its tract (through the hyoid bone, sometimes found passing above or below the hyoid) was freely released from surrounding muscular structures. Dissection continued deeply to include the tract of the TGDC and a core of tongue musculature to the level of lingual mucosa where the tract was transected.

5. The tongue defect was tightly closed and the remnant strap muscles were approximated. A Jackson-Pratt drain was inserted when indicated, and the wound was closed layer by layer.

Evaluation of the non-fusion cartilaginous portion of the hyoid

In case of suspected TGDC, we checked for non-fusion cartilaginous portions at both sides of hyoid body by preoperative neck CT scan. Preoperative neck CT is a relatively accessible modality. Despite the utility of preoperative ultrasonography that is commonly used in the workup of a midline neck mass in the pediatric population, the diagnostic accuracy of this imaging modality may have significant limitations. [16] Therefore, most TGDC patients undergo a preoperative work-up that includes neck CT. We routinely perform neck CT as a contrast-enhanced protocol using a General Electric Light Speed Ultra Scanner (General Electric Medical Systems, Milwaukee, WI, USA) to rule out other tumorous condition including dermoid cyst, ectopic thyroid cyst, and rarely malignant TGDC, and to determined more precisely the features of the mass. We have used the following scanning parameters: a 2 × 0.625-mm detector configuration; 2.5–3.0 mm slice thickness; 2.5–3.0 mm interval; 200–250 mA; 120kVp; and a 512 matrix. Evaluation of the scans was performed using PACS workstation software (PACS; Marotech, Seoul, Korea).

The degrees of fusion at the cartilaginous portion of the hyoid on axial CT views were classified as follows, based on a revised concept of classification adopted from previous publication[17] (Figure 3):

(1) Non-fusion: no focal fusion was detected at the area of contact between body and cornu. The line of the gap between body and cornu was definite over the full length of contact, regardless of the breadth of the gap; (2) Partial fusion: the line of fusion was advanced to involve<3/4 of the contact line; (3) Complete fusion: fusion

of>3/4 of the contact line.

Comparison between hyoid cartilage division and conventional bone cutting and statistical analysis

To compare and evaluate the feasibility of the operative techniques, the retrospective chart review was undertaken with respect to age, sex, operation time, postoperative drain insertion, amount of drainage, length of postoperative hospitalization, other operation-related complications and recurrence. Data were analyzed with Statistical Program for Social Sciences (SPSS) for Windows version 20.0 software (Chicago, IL). Simple descriptive statistics were used to describe the patient population. The Mann–Whitney test and Fisher's exact test were used for comparison, and statistical significance was accepted at $p<0.05$.

Results

Among59 patients, 27 pediatric (under 15 years) patients (median 6years; range, 1–13 years) and 32 adult patients (median 37years; range, 16–66 years) were included in the current study. The mean follow-up duration was 61.7± 31.3 months and a recurrent TGDC was re-excised at post-operative 18 months in only one pediatric case. There were no specific complications such as postoperative fistula formation or postoperative infection.

Among the pediatric patients, 96% (26/27) showed non-fusion of the hyoid, and all of these patients were treated with the cartilage division method. Only one pediatric patient, who had a partial fusion of the hyoid, was treated by a conventional hyoid bone cutting method. Other patient characteristics are summarized in Table 1. Almost all cases of TGDC showed bilateral patterns of hyoid fusion. However, there were 3 cases of asymmetric fusion among the adult patients, in which one side showed complete fusion and the other side showed partial fusion. All of these cases were treated with a conventional bone cutting method.

Among the 32 adult TGDC cases, there were 13 (41%) complete fusion cases, 5 of which were treated with the cartilage division method. Seven (22%) adult patients had non-fusion of the hyoid on CT scan, and all of these were successfully by the cartilage division method. Eight of the 12 remaining adult patients, who had focal or partial fusion, were also treated with the cartilage division method. Therefore, a total 20 of 32adult patients (63%) with TGDC were treated with the cartilage division method. The ages (44.8± 14.9) of adult patients treated with hyoid cartilage division methods ranged from 16 to 66 years and were not statistically different

from the ages (36.2± 15.8) of the adult TGDC patients who were treated with a conventional bone cutting method (Mann–Whitney U test, p=0.139).

Statistical comparisons between operative techniques were performed for the adult TGDC cases in order to examine of feasibility of the hyoid cartilage division method in adults, and are summarized in Table 2. Patients who were treated with the cartilage division methods showed better results in all parameters, including operative time, length of hospitalization, and amount of drainage. However, there were no statistical differences in non-parametrical analysis (Mann–Whitney U test and Fisher's Exact test). The postoperative outcomes in pediatric patients are described in Table 3.

Discussion

The classical "core principle" originated from Schlange's initial concept of removing the middle third of the hyoid bone in 1983. Later, Sistrunk extended this dissection to the foramen caecum at the base of tongue and reported reduced recurrences rates and improved surgical results of TGDC excision. These concepts retain the general principles of the classic Sistrunk procedure and this procedure remains the gold standard for surgical management of TGDC, with extensive support in the literature to this day.[2, 12, 13, 18] The hyoid cartilage division method described in this study is an adapted Sistrunk operation with a modified procedure for central hyoid removal, and the "core principle" is still applied.

In the conventional Sistrunk operation, the central body or middle third of the hyoid is transected on both sides of body of the hyoid, about 1/4 inch in length, with bone-cutting forceps. Although bleeding from soft tissues can easily controlled by cauterization, which causes micro vessels to collapse and become sealed, bone bleeding, as usually occurs through channels in the bony cancellous structures at the transected hyoid body, and requires a meticulous hemostasis with thorough electro-cauterization and additional bone wax. The method of cartilage division using monopolar Bovie electro-cauterization can avoid unnecessary bone bleeding. In case of non-fused hyoid, the end of the hyoid body can easily be separated from the synovial joint by traction to the hyoid and monopolar dissection of dense fibrous strips at the cartilaginous junction between the body and the greater horn. Further, since an immaturely ossified body of the hyoid is shorter than a fully ossified hyoid bone, it should not

be necessary to dissect the attached strap muscles along the full length of the hyoid to gain approach to the cartilaginous portion of the hyoid. Therefore, bleeding from the dissected muscles can also be minimized during the hyoid cartilage division technique (Figure 2).

The ossification process of the hyoid proceeds through fusion of ossifying centers of the body and greater and lesser cornua of the hyoid. The age of fusion of the greater cornu with the body of the hyoid has been well-examined in published forensic studies and covers a diverse range from 30–40 years to 60 years.[14, 17, 19, 20] In our results, therefore, the body of the hyoid bone could be separated from the greater cornu without bone cutting or fracture and then simply divided by cartilage division method during TGDC operations even in adults. In cases of non-fused hyoid detected on preoperative neck CT scan, especially in pediatric patients, the cartilage division was more easily achieved.

To diagnose the pediatric neck mass, ultrasound (US) has been regarded as the first modality for investigation, as it is readily available and does not involve ionizing radiation, which computed tomography (CT) has, and does not require the sedation of infants as often as magnetic resonance imaging (MRI).[21] Due to the low cost for CT in Korea, CT scans are used indiscriminately as an initial work-up for pediatric patients.[22] Since US requires training for performing US examination and interpretation of the results, less-experienced physicians might first order CT scans for even small neck lesions.[23] It could result in omitting the evaluation by US. Therefore the majority of patients who included this study already have taken CT before the first visiting to this tertiary hospital. But as you can see from our results, CT is not essential for confirming the non-fused hyoid. Especially in pediatric patients, US is enough for diagnosis and treatment TGDC.

Owing to the statistical limitations of our study, comparison of parameters related to the operative results between two operative techniques was performed only for adult cases. However, we could assure that our procedure using the cartilage division method is as feasible and safe as the conventional bone cutting methods. The postoperative results, including operation time, length of hospitalization, and amount of drainage were slightly better in hyoid cartilage division cases compared to conventional methods. To further validate the procedure, future randomized controlled studies using both the cartilage division technique and conventional procedures should be performed.

Conclusion

During Sistrunk operation for TGDCs, our hyoid cartilage division technique is feasible and comparable to conventional hyoid bone cutting in terms of postoperative results. Our results suggest that our procedure may be of value for patients with TGDC in the presence of hyoid on-fusion, as in most pediatric and some adult patients, in that it may minimize the risk of bleeding, the length of hospitalization, and the drainage amount.

References

[1] Allard RH. The thyroglossal cyst. Head & neck surgery 1982; 5:134-146.

[2] Shah R, Gow K, Sobol SE. Outcome of thyroglossal duct cyst excision is independent of presenting age or symptomatology. International journal of pediatric otorhinolaryngology 2007; 71:1731-1735.

[3] Ewing CA, Kornblut A, Greeley C, Manz H. Presentations of thyroglossal duct cysts in adults. Eur Arch Otorhinolaryngol 1999; 256:136-138.

[4] Kurt A, Ortug C, Aydar Y, Ortug G. An incidence study on thyroglossal duct cysts in adults. Saudi medical journal 2007; 28:593-597.

[5] Sprinzl GM, Koebke J, Wimmers-Klick J *et al.* Morphology of the human thyroglossal tract: a histologic and macroscopic study in infants and children. The Annals of otology, rhinology, and laryngology 2000; 109:1135-1139.

[6] Sammarco GJ, Mc Kenna J. Thyroglossal duct cysts in the elderly. Geriatrics 1970; 25:98-101.

[7] Brousseau VJ, Solares CA, Xu M *et al.* Thyroglossal duct cysts: presentation and management in children versus adults. International journal of pediatric otorhinolaryngology 2003; 67:1285-1290.

[8] Burkart CM, Richter GT, Rutter MJ, Myer CM, 3rd. Update on endoscopic management of lingual thyroglossal duct cysts. The Laryngoscope 2009; 119:2055-2060.

[9] Byeon HK, Ban MJ, Lee JM *et al.* Robot-assisted Sistrunk's operation, total thyroidectomy, and neck dissection via a transaxillary and retroauricular (TARA) approach in papillary carcinoma arising in thyroglossal duct cyst and thyroid gland. Annals of surgical oncology 2012; 19:4259-4261.

[10] Kayhan FT, Kaya KH, Koc AK *et al.* Transoral surgery for an infant thyroglossal duct cyst. International journal of pediatric otorhinolaryngology 2013; 77:1620-1623.

[11] Kim CH, Byeon HK, Shin YS *et al.* Robot-assisted sistrunk's operation via a retroauricular approach for thyroglossal duct cyst. Head & neck 2013.

[12] Chou J, Walters A, Hage R *et al.* Thyroglossal duct cysts: anatomy, embryology and treatment. Surgical and radiologic anatomy : SRA 2013; 35:875-881.

[13] Sistrunk WE. The Surgical Treatment of Cysts of the Thyroglossal Tract. Annals of surgery 1920; 71:121-122 122.

[14] Gupta A, Kohli A, Aggarwal NK, Banerjee KK. Study of age of fusion of hyoid bone. Leg Med (Tokyo) 2008; 10:253-256.

[15] Urbanova P, Hejna P, Zatopkova L, Safr M. The morphology of human hyoid bone in relation to sex, age and body proportions. Homo : internationale Zeitschrift fur die vergleichende Forschung am Menschen 2013; 64:190-204.

[16] Sidell DR, Shapiro NL. Diagnostic accuracy of ultrasonography for midline neck masses in children. Otolaryngology--head and neck surgery : official journal of American Academy of

Otolaryngology-Head and Neck Surgery 2011; 144:431-434.
[17] Harjeet K, Synghal S, Kaur G *et al.* Time of fusion of greater cornu with body of hyoid bone in Northwest Indians. Leg Med (Tokyo) 2010; 12:223-227.
[18] Hussain K, Henney S, Tzifa K. A ten-year experience of thyroglossal duct cyst surgery in children. Eur Arch Otorhinolaryngol 2013; 270:2959-2961.
[19] Miller KW, Walker PL, O'Halloran RL. Age and sex-related variation in hyoid bone morphology. Journal of forensic sciences 1998; 43:1138-1143.
[20] Parsons FG. The Topography and Morphology of the Human Hyoid Bone. Journal of anatomy and physiology 1909; 43:279-290.
[21] Faerber EN, Swartz JD. Imaging of neck masses in infants and children. Critical reviews in diagnostic imaging 1991; 31:283-314.
[22] Lee DY, Seok J, Kim YJ *et al.* Neck computed tomography in pediatric neck mass as initial evaluation in ED: is it malpractice? The American journal of emergency medicine 2014; 32:1237-1240.
[23] Collins B, Stoner JA, Digoy GP. Benefits of ultrasound vs. computed tomography in the diagnosis of pediatric lateral neck abscesses. International journal of pediatric otorhinolaryngology 2014; 78:423-426.

Table 1.Demographic characteristics of the patients with thyroglossal duct cysts (TGDCs)

	Pediatric TGDCs (N= 27)		Adult TGDCs (N=32)	
Age (Median, range, years)	6(1 – 13)		37 (16 – 66)	
Sex (Male : Female)	22 : 5		17 : 15	
Operative Technique				
Conventional Hyoid Cutting	1 (4%)		12 (37%)	
Hyoid Cartilage Division	26 (96%)		20 (63%)	
Hyoid Fusion Classification & the proportion of Hyoid cartilage division in each classification				
	Frequency	Cartilage division	Frequency	Cartilage division
I. Non-Fusion	26 (96%)	26/26	7 (22%)	7/7
II. Partial Fusion	1 (4%)	0/1	12 (37%)	8/12
IV. Complete Fusion	0	0/0	13† (41%)	5/13
Follow-up (month)	62.8± 29.5		58.1±32.2	
Op-relatedcomplication	0		0	
Recurrence	1		0	

† Three asymmetric fusion cases were included.

Table 2.Comparison of postoperative results between operative techniques in adultthyroglossal duct cyst (TGDC) patients

	Cartilage Division (N=20)	Conventional Bone Cutting (N=12)	*p*-value
Age(years)	44.8± 14.9 (16 - 66)	36.2± 15.8 (18 – 66)	0.139
Operation Time (min)	61.3±24.6	66.0 ±31.0	0.480
Length of Hospitalization(days)	2.8 ± 0.8	4.1± 2.7	0.089
Drain Insertion (cases)	9 (45%)	8 (66.7%)	0.291
Drainage Amount (mL)	45.9± 31.3	57.8± 46.4	0.336

Table 3.Comparison of postoperative results between operative techniques in pediatric thyroglossal duct cyst (TGDC) patients

	Cartilage Division (N=26)	Conventional Bone Cutting (N=1)
Age (years)	5.8 ± 14.9 (1 - 13)	10
Operation Time (min)	62.3±29.2	55
Length of Hospitalization (days)	2.8 ± 0.8	3
Drain Insertion (cases)	5 (19%)	1
Drainage Amount (mL)	38.2 ± 22.4	50

Figure 1. Scheme of Sistrunk operation. A. Traditional Sistrunk operation. B. Modified Sistrunk operation: division of hyoid cartilage

Figure 2. Excision of TGDC with modified technique. * Divided cartilage portion of hyoid bone

Figure 3. Classification of cartilage fusion of hyoid. A. Non-fusion, B. Partial fusion, C. Complete fusion

Figure(s)