

Do We Really Need to Perform the Original Sistrunk Operation When Treating Thyroglossal Duct Cysts?

(Tiroglossal duktus kisti tedavisinde orjinal sistrunk ameliyatı gerçekten gerekli mi?)

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ÖZET

Amaç: Tiroglossal duktus kisti (TGDK) çocuklarda sık görülen bir konjenital boyun anomalisidir. Tedavisi zamanla bazı modifikasyonlara uğramış olan ve 1920 de Sistrunk tarafından tariflenen şekliyle cerrahi eksizyondur. Bu çalışmayla biz kliniğimizde TGDK veya fistülü nedeniyle opere edilen hastaların klinik özelliklerini ve tedavi sonuçlarını değerlendirmeyi amaçladık.

Materyal ve Metot: 2005 ve 2012 yılları arasında boyun orta hatta şişlik ön tanısıyla kliniğimizde tedavi edilen 48 çocuk hastanın geriye dönük olarak kayıtları incelendi. Patoloji sonuçları neticesinde TGDK kesin tanısı alan 34 hasta çalışmaya dahil edildi.

Bulgular: Hastaların 17'si erkek (%50) ve 17'si kızdı (%50). Ortanca yaş 5,4 yıl olarak bulundu. Hastaların 17'sinin (%50) tedavisinde klasik Sistrunk operasyonu uygulanmış olmasına rağmen, 17'sinde (%50) operasyon Schlange'nin tariflediği şekilde hiyoid kemik korpusu eksizyonu sonrası ilave disseksiyon yapılmadan sonlandırılmıştı. Ortalama 20,5 ay takip sonrasında opere edilen hastaların hiçbirisinde nüks tespit edilmedi.

Tartışma: Hastalarımızın %50'sinin tedavisinde Schlange prosedürü uygulamamıza rağmen nüks saptamadık. Bu çalışma hiyoid kemik korpusunun eksizyonunun TGDK tedavisinde temel prensip olduğunu göstermektedir.

Anahtar Kelimeler: Tiroglossal kanal kisti; çocuk; sistrunk ameliyatı; schlange ameliyatı

ABSTRACT

Background: Thyroglossal duct cyst (TDC) is a common congenital midline neck anomaly among children. Although the widely excepted treatment of TDC is surgical excision as described by Sistrunk in 1920, some modifications have been implemented by the time. In this study we aimed to evaluate the clinical features and treatment results of the patients operated on TDCs in our department.

Material and Methods: A retrospective review of 48 patients who had been operated for midline neck swelling in our department between January 2005 and February 2013 was conducted. Thirty-four patients with histopathological diagnosis of TDC were enrolled to this study.

Results: Of the patients, 17 (50%) were male and 17 (50%) were female and the median age of the patients was 5.4 years. Though the classical Sistrunk operation was performed on 17 patients (50%), we realized that in the remaining 17 (50%) patients, the dissection was finalized immediately after the excision of hyoid bone corpus as described by Schlange. None of the patients operated for TDC had recurrent disease during a median follow-up period of 20.5 months.

Conclusions: Although Schlange procedure was performed on 50% of the patients, we did not encounter any recurrences. Thus this study demonstrates that hyoid bone corpus excision is the principal component of TDC treatment.

Key Words: Thyroglossal duct cyst; children; sistrunk procedure; schlange procedure

INTRODUCTION

Thyroglossal duct cyst (TDC) is a common congenital midline neck anomaly among children and occurs along the path of thyroid descent from the foramen cecum to the lower neck¹. Though, it is commonly determined as a painless swelling in

the midline, sometimes emerges with infection which contributes to recurrence of the disease.

Today, widely accepted treatment of the disease is surgical excision including complete excision of the cyst and its tract with hyoid bone corpus removal up to the base of the tongue as described by Sistrunk in 1920. Since Sistrunk has introduced this technique, the recurrence rate of the disease significantly decreased². By the time some modifications to this technique have been implemented and still recurrence of the disease is the most common problem. In this study we aimed to evaluate the clinical features and

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treatment results of the patients operated on TDCs.

MATERIAL and METHODS

After obtaining institutional ethic committee approval, a retrospective review of 48 patients who had been operated for midline neck swelling in our department between January 2005 and February 2013 was conducted in our department. Patient charts including detailed operative notes and histopathological results of specimens were investigated. Thirty-four patients with histopathological diagnosis of TDC were enrolled to this study. Patient demographics, clinical characteristics, treatment modalities and follow-up results were assessed.

Statistical analysis was done using SPSS software, version 15.0 (SPSS Inc, Chicago, IL, USA). Data were reported as median and range. Chi-square test and Fisher's exact test were used for categorical variables and Mann-Whitney U test was used for continuous variables. A p value $< .05$ was considered as significant.

RESULTS

Of the patients, 17 (50%) were male and 17 (50%) were female. Median age of the patients was 5.4 years (ranging from 5 months to 13 years). Complaints of the patients during initial admission were swelling on neck in 24 (71%), infected swelling in 9 (26%) and fistula orifice in one (3%) patient. Two patients with infected swelling on the neck were previously operated for neck cyst in other centers. While the cystic mass was mobile with swallowing within 28 (82%) patients, the cystic mass was immobile in 6 (18%) of the patients. TDC was localized in the midline in 25 (73%), slightly on the right side of midline in 6 (18%) and slightly on the left side of the midline

in 3 (9%) of the patients. Additionally, TDCs were infra hyoid in 20 (59%) and on the hyoid bone in 14 (41%) of the patients. Neck US was performed on 27 (79%) of the patients, one patient required MRI evaluation because of solid mass on US and Tc-99m pertechnetate thyroid scintigraphy was performed on 5 patients subsequent to increased TSH values in thyroid function tests to exhibit thyroid gland abnormalities, but none of the evaluations revealed an ectopic thyroid tissue.

Prior to surgery 13 (38%) patients had infected TDC and antibiotic treatment was implemented during preoperative course and 6 (18%) patients required incision and drainage. None of the patients had family history of TDC. Three patients had additional anomalies including Hirschsprung disease, undescended testis and facial hemangioma.

Surgical procedures were assessed via the operative notes and histopathological examination reports of the specimens. Though the classical Sistrunk operation was performed on 17 patients (50%), we realized that in the remaining 17 (50%) patients dissection up to the base of tongue was not performed and finalized immediately after the excision of hyoid bone corpus as described by Schlange. None of the patients operated for TDC had recurrent disease during a median follow-up period of 20.5 months (ranging from 1 to 85 months). Subsequently we have considered the clinical features of the two operation types (Table 1). There was no statistically significant difference in terms of gender, median age, preoperative infection, TDC localizations, movement with swallowing, operative time or follow-up periods ($p>0.05$). We did not encounter any postoperative complications or recurrences due to surgical procedures.

Table 1. Consideration of two Operation Types According to Demographic and Clinical Features of the Patients

		Sistrunk procedure	Schlange procedure	p value
Gender (Male/Female)		6/11	11/6	0.169
Median Age (years)		4.7 (5 months-12.5 years)	6.2 (16 months-13 years)	0.185
Preoperative infection (n,%)		5 (29%)	8 (47%)	0.481
Movement with swallowing (n,%)		15 (88%)	13 (77%)	0.656
Localization according to midline (n,%)	Slightly right	3 (18%)	3 (18%)	0.830
	Slightly left	2 (12%)	1 (6%)	
	Midline	12 (70%)	13 (76%)	
Localization according to hyoid bone (n,%)	Hyoid	6 (35%)	8 (47%)	0.728
	Infra hyoid	11 (65%)	9 (53%)	
Operation time (minutes)		70 (26-120)	50 (30-108)	0.162
Follow-up period (months)		16 (1-66)	25 (1-85)	0.756

DISCUSSION

TDC is the most common congenital midline neck anomaly among children¹. TDC is frequently adjoins to hyoid bone and begins from foramen cecum, and it may emerge from anywhere on the descent path of thyroid gland. The clinical signs of TDC may be a painless midline swelling, a purulent discharging fistula or an infected tender mass on the neck². Although the movement of cystic mass with swallowing or with protrusion of the tongue is usually enough for the diagnosis; dermoid cyst, branchial cleft cyst, cystic hygroma, lipoma, sebaceous cyst or ectopic thyroid tissue should be considered in the differential diagnosis of TDC³. Of the masses that are considered as TDC, 1-2% may be midline ectopic thyroid tissue¹. It is crucial to exclude an ectopic thyroid gland with preoperative laboratory and imaging studies¹. Although, serum TSH value and demonstration of a thyroid gland with normal morphology on US is usually sufficient, in case of an increased TSH value with hypothyroidism signs and absence of a normal thyroid gland on US, performing a thyroid scintigraphy has been recommended⁴.

Avoiding complications due to recurrent infections and risk of malignancy, though it is a rare occasion, the treatment of TDC is surgical excision. During embryological development, thyroglossal duct passes through anterior, posterior or commonly inside of the hyoid bone and frequently branches into small canals at this level^{5,6}. Until the relationship between thyroglossal duct and hyoid bone has clarified, the recurrence rate up to 50% has decreased to 20% when Schlange proposed excision of hyoid bone corpus with TDC, for the first time in 1893. Afterwards in 1920 Sistrunk suggested excision of proximal dissection of the duct structures reaching up to the foramen cecum in addition to hyoid bone corpus excision and reduced the recurrence rate to 3-4%^{2,3,5}. Today, although Sistrunk operation is the gold standard for the treatment of TDC, still high recurrence rates up to 10% have been reported¹. Incomplete excision of thyroglossal duct, postoperative infections, intraoperative

rupture of the cyst and incomplete excision of hyoid bone corpus have been proposed to associate with recurrence^{7,8}.

By the time, the original operation defined by Sistrunk has been modified by some authors. Horisawa et al. have suggested that after excision of hyoid bone corpus 5mm more proximal dissection and excision of the tract would be sufficient especially among children aged 2-6 years. Based on the 3-dimensional reconstruction of the TDC specimens they indicated that the thyroglossal duct remnants converts into a single channel at this level and at the level of tongue base thyroglossal duct again branches into multiple channels and secretory glands thus they have proposed that the dissection extending to foramen cecum would be insufficient for excising all these branches⁵. On the other hand, various studies indicate that the proximal dissection should be completed as close as possible to the foramen cecum to prevent recurrences, as well as, wider excision of hyoid bone than Sistrunk had defined has been suggested^{2,6}.

CONCLUSIONS

Of the patients in this study, proximal dissection was not performed in 50% following hyoid bone corpus excision. Actually the excision of TDC in these patients was finalized with hyoid bone corpus removal as defined by Schlange and during 25,5 months median follow-up period any recurrence was encountered among these patients. Additionally, during the preoperative course proper treatment of infected TDCs, which have significant importance on recurrences, might be effective on postoperative uneventful period among these patients. Though this study demonstrates the importance of hyoid bone corpus excision, to discuss the necessity of proximal dissection extending to foramen cecum, the number of patients in this study is insufficient and requires larger series.

Conflict of interest statement: None declared.

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