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Possible Risk Factors for Recurrence After Removal of Thyroglossal Duct Cysts with the Sistrunk Procedure

Sistrunk Prosedürü Sonrası Tiroglossal Kanal Kisti Nüksleri İçin Olası Risk Faktörleri

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ABSTRACT Objective: To evaluate the risk factors except surgery that may affect the recurrence rate of thyroglossal duct cysts (TGDCs). **Material and Methods:** The patients who had undergone surgery for TGDCs between 2009 and 2019 were examined. Cases operated with a technique other than the Sistrunk procedure and the revision cases were excluded from the study. Age and gender distributions, complaints, number of infectious attacks, presence of a fistula, radiological imaging, mass characteristics, surgical findings, hospitalization time, pathological examination results, and presence of complications were assessed. The effects of these parameters on the recurrence were investigated. **Results:** Fifty-eight patients were included in the study (43.1%, 25 females and 56.9%, 33 males). The mean age was 21.53±19.19 years (range: 2-78 years). The most common complaint was neck mass (n=49, 84.5%). Fifteen patients (25.9%) had a fistulized skin lesion. The most common location was the infrahyoid region (46.6%). The recurrence rate was found to be 13.8% (8/58). The mean recurrence time was 1.8 years. In two patients' pathological examination revealed papillary thyroid cancer (3.4%). The recurrence rates in the patients who had pain, fistulized skin lesions, and masses located above the hyoid level were found to be statistically significant (p=0.01, p<0.01, p=0.03, respectively). **Conclusion:** Location of the mass, presence of pain, and fistulized skin lesions are the factors affecting the recurrence in the patients undergoing the Sistrunk procedure.

Keywords: Thyroglossal duct; cyst; fistula; recurrence; the Sistrunk procedure

ÖZET Amaç: Tiroglossal kanal kistleri [thyroglossal duct cysts (TGDC)]nin nüks oranını etkileyebilecek cerrahi yöntem dışı risk faktörlerini değerlendirmek. **Gereç ve Yöntemler:** 2009-2019 yılları arasında, TGDC sebebiyle ameliyat edilen hastalar incelendi. Sistrunk prosedürü dışında bir teknikle opere edilen olgular ve revizyon olguları çalışma dışı bırakıldı. Yaş ve cinsiyet dağılımları, semptomlar, enfeksiyon atağı sayısı, fistül varlığı, radyolojik görüntüleme, kitle özellikleri, cerrahi bulgular, hastanede yatış süresi, patolojik muayene sonuçları ve komplikasyon varlığı değerlendirildi. Bu parametrelerin, nüks üzerine etkileri araştırıldı. **Bulgular:** Elli sekiz hasta çalışmaya dâhil edildi (25 kadın %43,1; 33 erkek %56,9). Ortalama yaş, 21,53±19,19 (dağılım: 2-78) yıl idi. En sık şikâyet (n=49; %84,5) boyun kitlesi idi. On beş (%25,9) hastada fistülize cilt lezyonu vardı. En sık yerleşim yeri (%46,6), infrahyoid bölge idi. Nüks oranı %13,8 (8/58) olarak bulundu. Ortalama nüks süresi 1,8 yıldı. İki hastanın patolojik incelemesinde , papiller tiroid kanseri (%3,4) saptandı. Ağrı, fistülize cilt lezyonları ve hiyoid düzeyinin üzerinde yerleşimli kitle bulunan hastalarda, nüks oranları istatistiksel olarak anlamlı şekilde daha yüksek bulundu (sırasıyla p=0,01; p<0,01; p=0,03). **Sonuç:** Kitlenin yeri, ağrı varlığı ve fistülize cilt lezyonları, Sistrunk prosedürüne tabi tutulan hastalarda nüksü etkileyen faktörlerdir.

Anahtar Kelimeler: Tiroglossal kanal; kist; fistül; nüks; Sistrunk prosedürü

Thyroglossal duct cysts (TGDCs) are the most common congenital cystic neck masses in the head and neck regions and occur in approximately 7% of the population.[1] Thyroglossal duct cysts may arise anywhere on the embryological migration pathway of the thyroid gland, that is, between the foramen cecum and inferior central neck. The cysts develop from secretions of epithelial remnants if the thy-

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roglossal duct is not completely closed. In general, a TGDC exists as a painless and mobile mass in the midline of the neck in childhood.[2] The movement of the mass during swallowing and tongue movements is an important diagnostic feature.[1,2] It may also exist as abscess or fistulae formation in the skin. In the case of an acute infection, the operation is not recommended unless the infection is controlled with an appropriate antibiotherapy.[2]Ultrasonography (USG) is the first-choice imaging modality to confirm the diagnosis and to investigate the presence of ectopic thyroid tissue.[3] Histopathologic examination reveals malignancy in approximately 1% of TGDC patients operated. The most common malignancy is papillary thyroid carcinoma that accounts for 80% of these cases.[4] The main treatment modality is the Sistrunk procedure that includes complete removal of the cyst with the corpus of hyoid bone and cyst tract.[5] Recurrence is the most common complication and the major factor affecting the recurrence is an inadequate surgery. In the literature, the recurrence rates for the simple cyst excision and the Sistrunk procedure were reported to be around 40% and 5%, respectively.[1,6] The only generally-accepted risk factor credited with causing the cyst recurrence is an inappropriate surgical method.[7,8] The aim of this study was to evaluate the possible risk factors affecting the recurrence after removal of thyroglossal duct cysts with the Sistrunk Procedure.

MATERIAL AND METHODS

Fifty-eight patients who had undergone an operation for a thyroglossal cyst or a fistula between 2009 and 2019 were included in the study. The subjects operated with a technique other than the Sistrunk procedure were excluded from the study. The Sistrunk procedure involves complete removal of the cyst with the corpus of hyoid bone and cyst tract (Figure 1). Moreover, revision cases were excluded from the study in order to investigate the relationship between the lesion characteristics and the recurrence. Patient files were retrospectively reviewed. The parameters including age and gender distributions, complaints, number of infectious attacks, presence of a fistula, radiological imaging, mass characteristics, surgical findings, hospitalization time, pathological examination results, and presence of complications and recurrence were assessed. The mean follow-up period was 5.2 years. All the patients underwent preoperative imaging. Ultrasonography, magnetic resonance imaging (MRI), and computed tomography (CT) were each the imaging modality used in 45, 6, and 7 patients, respectively. Imaging methods identified a thyroglossal duct cyst as a preliminary diagnosis in all the patients. Therefore, diagnostic fine-needle aspiration cytology was not considered necessary in most cases. Statistical analysis was performed using the SPSS v22 (IBM, New York, NY, USA) program. Descriptive statistics of the demographic data were calculated. Chi-square tables were used to compare the recurrence rates in different groups.

FIGURE 1: The Sistrunk operation for a thyroglossal duct cyst. Removal of the whole of the cyst and corpus of hyoid bone.

All investigations were performed in accordance with the Declaration of Helsinki on biomedical studies involving human subjects, and informed consent was obtained from all the study subjects. The study was approved by the local Institutional Review Board of Gazi University with protocol number: 24.10.2019-E.133049.

RESULTS

Of 58 patients included in the study, 56.9% (33) were male and 43.1% (25) were female. The range of the patients' age was 2-78 years with a mean value of 21.53 ± 19.19 years. The disease most-commonly emerged in the first or second decade. The most common presenting complaint was neck mass (n = 49, 84.5%). Apart from the mass complaint, five patients (8.6%) presented with inflammatory discharge, three (5.2%) with dysphagia, and one (1.7%) with hoarse-

ness. In addition, 21 patients (36.2%) suffered from pain. Forty-three patients operated had an isolated cyst while 15 patients (25.9%) had a fistula formation in the skin. The mean time duration elapsed after the diagnosis was 29.60 ± 49.90 months (range: 1 month to 20 years). Twenty-nine patients had an infection attack at least once. It was observed that 10 patients had one attack. One of the patients had more than 10 attacks. According to lesion location, 27 patients (46.6%) had infrahyoid localization, the lesion was above the hyoid level in 17 patients (29.3%), and 14 patients (24.1%) had a lesion located at the hyoid level (Table 1).

Recurrence was observed in eight patients during follow-up (13.8%). The mean recurrence period was 1.8 year. Seroma and wound infection were observed in three patients in the early postoperative period (5.1%). There were no other serious complications. The duration of hospitalization ranged from 1 to 6 days with a mean value of 2.48 ± 1.20 days. The cyst was smaller than 3 cm in 26 patients (44.8%) and larger in 32 patients (52.2%).

All patients (n=58) had undergone the Sistrunk operation as a surgical procedure. Pathological examination revealed papillary thyroid cancer in 2 patients (3.4%) (Table 1).

Of the patients with recurrence (8 patients), six were male and two were female. However, no statistically-significant effect of the gender on the recurrence was observed. There was no significant difference between decades in terms of the recurrence frequency. The duration of symptoms and the number of infectious attacks also had no statistically-significant effect on the recurrence. However, six of the patients with recurrence had at least one history of infection attacks. Recurrence was more frequent in patients with pain at presentation (p=0.01). The recurrence rates of the patients who had fistulized skin lesions on admission were higher than those of the patients who presented with cysts. (p<0.01). TGDCs located above the hyoid level also had higher recurrence rates than those of TGDCs located at or below the hyoid level. (p=0.03) There was no significant effect of the mass size on the recurrence (Table 2).

DISCUSSION

Thyroglossal duct cysts (TGDCs) develop as a result of the inadequate obliteration of the thyroglossal duct that occurs in the embryological migration pathway of the thyroid gland. The collection caused by epithelial secretions within the duct creates a cyst formation and the cyst then becomes symptomatic.[2] It is often seen in the midline and exists as a neck mass. In our series, in accordance with the literature the most common presentation was an asymptomatic midline neck mass, which was the first finding in 49 patients (84.5%).[7] It may exist as a cyst or a fistula. In our case series, 15 patients (25.9%) presented with fistula. This ratio is similar to that of the report by Rohof et al. in which 50 out of 207 patients presented with a fistula.[6] History and physical examination are the keys to diagnosis. In addition, imaging is helpful to confirm a diagnosis and thus, USG, CT, and MRI can be used to this end. The typical appearance of the lesion in USG is a thin-walled anechoic cyst. Unlike dermoid cysts, TGDCs have an irregular shape, heterogeneous internal echogenicity, and multilocularity in the USG images.[9] CT of the neck shows a hypodense mass with smooth borders. MRI helps to determine better the size and the borders of the mass, the invasion, and the malignancy. USG is an adequate and first-choice imaging method. CT is not preferred because of the ionizing radiation, the reasons why MRI is not preferred as a first-line imaging method include high cost, long procedure time, and the need for anesthesia.[10] In this study, USG was the most preferred imaging modality (45/58). We observed that the most common localization was in the infrahyoid region with a rate of 46.6%, followed by the suprahyoid location with 29.3%, and the hyoid level with 24.1%. The rates in our study are similar to the findings of Tristan et al.[11] Rohof et al. claimed the infrahyoid region (60.9%) to be the most common localization and found no statistical relationship between the recurrence and the localization.[6] In our study, a significantly high recurrence rate was observed in the lesions located in the suprahyoid region. The surgical technique is the most important factor that determines the recurrence in TGDCs. In cases with simple excision, the recurrence rate reaches 50%

TABLE 1: Demographic distribution of patients and characteristics of the mass.

Patient Characteristics	Gender	33/25(M/F)	56,9% / 43,1%
	Age	21,53	SD:19,19
	Mean Hospitalization (day)	2,48	SD:1,20
	Symptomatic Period (day)	29,6	SD:49,9
		(n)	**(%)**
Major Sign	Mass	49	84,5%
	Discharge	5	8,6%
	Dysphagia	3	5,2%
	Hoarseness	1	1,7%
Pain at Presentation	Yes	21	36,2%
	No	37	63,8%
Number of Infectious Attacks	0	29	50%
	1	10	17,2%
	2	9	15,5%
	3	4	6,9%
	4	3	5,2%
	6	2	3,4%
	>10	1	1,7%
		(n)	**(%)**
Imaging Technique	US	45	77,6%
	CT	7	12,1%
	MRI	6	10,3%
		(n)	**(%)**
Diagnosis	Cyst	43	74,1%
	Fistula	15	25,9%
		(n)	**(%)**
Location	Above the Hyoid	17	29,3%
	Hyoid	14	24,1%
	Sub-Hyoid	27	46,6%
		(n)	**(%)**
Recurrence	Yes	8	13,8%
	No	50	86,2%
		(n)	**(%)**
Early Postoperative Complication	Yes	3	5,2%
	No	55	94,8%
		(n)	**(%)**
Size	<3cm	26	44,8%
	>3cm	32	55,2%

while in the Sistrunk operation, the total removal of the mass with the middle part of the hyoid bone reduces the recurrence rate to 5% on average.[5,6,12]. In the literature, the recurrence rate varies between 0% and 15.8% in the Sistrunk operation series.[7,13-16] Marianowski R. et al had observed the recurrence rate to be 15.8% and concluded that the conditions related to the recurrence were revision cases, children being under 2 years, previous infections, and surgical methods other than the Sistrunk procedure.[16] Moreover, histopathologic confirmation in their study showed multiple extensions. Likewise, we encountered a relatively high recurrence rate in our study (13.8%). We concluded that the parame-

TABLE 2: Recurrence analysis.

		Recurrence			
		No	Yes	Total	p
Gender	Male	27	6	33	0,23
	Female	23	2	25	
Location	Sub-Hyoid	24	3	27	0,03
	Hyoid	14	0	14	
	Above the Hyoid	12	5	17	
Diagnosis	Fistula	8	7	15	<0,01
	Cyst	42	1	43	
Size	<3cm	22	4	26	0,52
	>3cm	28	4	32	
Pain	Yes	15	6	21	0.01
	No	35	2	37	
Number of Infectious Attacks	0	27	2	29	0,12
	>1	23	6	29	

ters related to the recurrence were the location of the mass, the presence of a fistula, and the pain complaints. The diagnostic error has been shown to be a factor affecting the recurrence.[17,18] This may be due to performing simple resection instead of the Sistrunk procedure in the patients without a diagnosis of TGDCs. In addition, the occurrence of cyst rupture during surgery has been associated with the recurrence.[1,15] The recurrence in TGDCs is often seen in the first year and the probability of recurrence significantly decreases after four years.[6] In our study, the mean recurrence time was 1 year and 10 months (range: 2 months-10 years). However, only two patients had a recurrence that occurred after one year. Studies have shown no association of the recurrence with patient age and sex (18-21). In our study, we did not observe any effect of age and gender on the recurrence rate.

In our study, no significant relation was found between the number of infection attacks and the recurrence. Similarly, Rohof et al. found no correlation between the infection and the recurrence.[6] However, there are also studies in the literature suggesting that infection increases the frequency of recurrence.[14-16,22,23] The use of antibiotics was not shown to prevent the recurrence.[14,23]

A significant difference was observed in the recurrence rates between primary and revision cases.[23,24] We examined the factors other than surgery affecting the recurrence rate by excluding revision cases from our study group and including only primary cases. In the present study, higher recurrence rates were observed in the patients who presented with local pain. This may be due to the relationship between fistulized skin lesions and pain. A statistically-significant high recurrence rate was observed in the patients with fistulized skin lesions. To our knowledge, this is the first study in the literature examining the relationship between pain and recurrence. Malignancy may rarely be encountered in the patients who undergo an operation due to TGDCs. This rate is approximately 1% in the literature data and 80% of these malignancies are papillary thyroid cancer.[4,25] Hurtle cell carcinoma, adenocarcinoma, anaplastic carcinoma, and non-Hodgkin's lymphoma have also been reported.[13] Taimisto et al. reported a malignancy rate of 3% in their series, all of which were papillary thyroid can-

cer.[12] In our study, malignancy was encountered in two patients (3.4%) and both were diagnosed as papillary thyroid carcinoma.

CONCLUSION

TGDCs should be considered when midline masses of the neck are encountered in both pediatric and adult populations. The gold standard treatment is surgery. The Sistrunk procedure is the main surgical procedure including complete excision of the cyst and its tract with the hyoid bone corpus. The most important factor determining the recurrence rate is whether an appropriate surgical modality was preferred. However, the recurrence rates in the patients undergoing the Sistrunk procedure suggest that there may be different factors other than surgery affecting the recurrence. The location of the mass, presence of pain, and fistulized skin lesions were found in this study to be the factors affecting the recurrence.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Muammer Melih Şahin;* ***Design:*** *Muammer Melih Şahin, Gökçen Cesur;* ***Control/Supervision:*** *Muammer Melih Şahin, Metin Yılmaz, Alper Ceylan;* ***Data Collection and/or Processing:*** *Muammer Melih Şahin, Süleyman Cebeci, Gökçen Cesur;* ***Analysis and/or Interpretation:*** *Muammer Melih Şahin, Recep Karamert, Mehmet Düzlü;* ***Literature Review:*** *Muammer Melih Şahin, Mehmet Düzlü, Recep Karamert;* ***Writing the Article:*** *Muammer Melih Şahin, Eray Uzunoğlu;* ***Critical Review:*** *Muammer Melih Şahin, Metin Yılmaz, Alper Ceylan.*

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