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Original Article

Diagnostic and Surgical Evaluation of Patients with Thyroglossal Duct Cysts and Fistulas: 7-Year Experience At Our Clinic

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ABSTRACT

Objective: Thyroglossal duct cysts (TGDCs) are one of the most common midline neck masses in children. They may be found in adults as well. The aim of this study was to evaluate the demographics of patients diagnosed with TGDCs and to discuss the diagnosis, treatment plans, and follow-up details.

Methods: The data of 91 patients diagnosed with TGDCs in our clinic between January 2010 and February 2017 were obtained. They included demographics, medical records, a postoperative follow-up, and complications. The pathology confirmed TGDCs in all 91 cases.

Results: Of 91 patients, 49 (53%) were males, and 42 (46%) were females. The mean age of patients was 20.29. Patients complained of a cystic midline mass in 47 (52%) of cases, and fistulas in the midline neck area in 43 of (47%) cases. All patients underwent the Sistrunk procedure. Fourteen (15%) patients relapsed.

Conclusion: TGDCs should be considered in differential diagnosis of midline neck masses in all ages. A physical examination and ultrasonography are the easiest and the most accurate methods in diagnosis. The Sistrunk procedure with its low recurrence rates is the gold standard method in the treatment.

INTRODUCTION

Thyroglossal ductus cysts (TGDCs) are the most common congenital midline neck masses, especially in the pediatric population. They are mostly seen in the pediatric age group, and approximately 50% of TGDCs are seen in the second decade of life. Furthermore, they can even be found later in adulthood.[1] They occur due to a failed regression of the thyroglossal duct, which is an embryonic remnant. The thyroglossal duct is found between the foramen caecum at the posterior portion of the tongue and the anatomical location of the thyroid gland. It involutes after five weeks of gestation.[2] Any defect in involution may result in a thyroglossal duct cyst or its remnant. Therefore, TGDCs may be found anywhere on this migrant tract. 60% of TGDCs are located in the thyroid region; 24% in the submental region; 13% in the substernal region; and 2% in the intralingual region.[3]

TGDCs make up the majority of congenital neck masses and are seen in 7% of the population.[4] In the pediatric population, they account for approximately 55%–75% of midline lesions.[5,6] The classical presentation is a painless, mobile, non-tender, and semisolid midline mass on the neck, which typically moves with swallowing. The cysts are usually 2–4 cm in size, but their size may increase during upper respiratory tract infections. They can make a fistula tract between the subcutaneous tissues and remnant ductus, a so-called thyroglossal fistula (TF). They may also contain the ectopic thyroid tissue. TGDCs are mostly benign in nature, but 1% may be malignant, and the most common malignancy is thyroid papillary carcinoma.[4,7]

In differential diagnosis, dermoid cyst, sebaceous cyst, lymphadenitis, lipoma, thyroid pyramidal lobe, and goiter should be kept in mind.[3] Ultrasonography (USG) is often sufficient to diagnose TGDCs and to distinguish ectopic thyroid tissue.[8] Computerized tomography (CT) and magnetic resonance imaging (MRI) can also be used for diagnosis. A fine needle aspiration biopsy (FNAB) may be occasionally required.

The main treatment method is surgery. The most common surgical method is the Sistrunk procedure. In this proce-

dure, TGDC is removed, including the central portion of the hyoid bone. The recurrence rate is less than 3% with this procedure when properly performed within 1 year.[5]

MATERIALS AND METHODS

This retrospective study was conducted using the electronic data of patients diagnosed with TGDC between January 2010 and February 2017, and it included 91 patients. The aim of this study was to evaluate patients who were diagnosed with TGDC and surgically treated in our clinic in terms of age, gender, time of onset of complaints, preoperative investigations, postoperative complications, and recurrence rates. Electronically collected data included patient demographics, medical records, and a postoperative follow-up.

The normal data distribution was tested with the Shaphiro–Wilk test. Student's t-test was used for the data of normal distribution of the variables, and the Mann–Whitney U test was used for those who did not show normal distribution. The relationships of two independent variables at the categorical measurement level were tested with a chi-squared test. The mean±standard deviation for numerical variables and the number and percentage values for categorical variables were given as descriptive statistics. The SPSS Windows package program was used for statistical analysis, and a p-value <0.05 was considered to be statistically significant.

RESULTS

A total of 91 patients were admitted to our outpatient clinic with a midline neck mass or fistula in the middle neck area. These patients were all evaluated clinically and underwent physical examination, 91 (100%) were evaluated with USG, 8 (8.8%) with a CT scan, 17 (18.7%) with MRI, and 31 (34.1%) additionally with FNAB. After a differential diagnosis of neck masses, all patients were diagnosed histopathologically with TDC or TF. Of 91 patients, 49 (53.8%) were males, and 42 (46.2%) were females (Table 1). The mean age was 20.29±15.61 years (Table 2).

The most common clinical finding was a mobile, semisolid, painless, cystic midline mass in 48 (52.7%) patients, whereas 43 (47.3%) presented with a fistula in the midline neck area. The movement of the mass with tongue protrusion was seen in all patients. All patients with TF complained of recurrent discharge from the fistula area. Patients with TGC had a history of recurrent swelling of the mass, but no discharge.

The Sistrunk procedure was applied to all patients. The cyst or fistula tract was excised with the corpus of the hyoid bone in all cases. In 14 (15.4%) patients, postoperative recurrence was observed. The average duration of recurrence was 4.93±7.16 years (Table 1).

Table 1. Patient demographics, characteristics of the mass, imaging modalities used in diagnosing, and recurrence rates after surgery

Variables	n	%
Gender		
Female	42	46.2
Male	49	53.8
Recurrent infection		
+	56	61.5
–	35	38.5
History of operation		
+	18	19.8
–	73	80.2
Fistula +/–		
+	43	47.3
–	48	52.7
Ultrasonography		
+	91	100
–	0	0
Computerized tomography		
+	8	8.8
–	83	91.2
Magnetic resonance imaging		
+	17	18.7
–	74	81.3
Fine needle aspiration biopsy		
+	31	34.1
–	60	65.9
Recurrence		
+	14	15.4
–	77	84.6

Table 2. Average age and symptom duration (years)

	Mean	Standard deviation
Age	20.29	15.61
Duration of symptoms	4.93	7.16

DISCUSSION

Thyroglossal duct anomalies result from a failed involution of the thyroglossal duct located between the foramen caecum at the base of the tongue and the thyroid gland. Therefore, a thyroglossal cyst may be seen at any place along this remnant duct.[2] In a study, thyroglossal duct cysts were in 60% of cases located adjacent to the hyoid bone, 24% between the hyoid bone and base of the tongue, 13% between the hyoid bone and the pyramidal lobe of the thyroid, and 3% were intralingual.[9] Sometimes, this cyst can get infected and cause a sinus between the cyst and the skin causing TF.

TGDCs make up the majority of congenital neck masses and are seen in 7% of the population.[4] Most commonly, they are found in the first decade of life. However, they can be also seen in adulthood. In our study, the mean age

of 91 patients was 20.29±15.61. The youngest patient was 3 years old. This study showed that mostly adult patients admitted to our clinic with a midline neck mass were diagnosed with TGDC. In a study, it was found that 53% of the patients diagnosed with TGDC were in the second decade, 24% in the third decade, and 23% over the age of 30.[10] The results from this study were similar to ours. This may be due to admittance of younger patients to pediatric/pediatric surgery outpatient clinics, or it may be due to lack of knowledge in differential diagnosis of midline neck masses.

In some studies, it was found that the male-to-female ratio is the same, whereas in some studies, there is male predominance.[2,5,6] In our study, 49 (53.8%) were males, and 42 (46.2%) were females, which shows male predominance.

Patients complain of recurrent swelling on the neck resolving with antibiotic treatment or other conservative measures. An acute airway obstruction, mass in the mouth floor, a severe infection of the mass and associated symptoms of infection, painful swelling and deglutination, dysphagia, dysphonia, etc. may also be among the symptoms. Moorthy et al.[11] found that 41.6% of their patients presented with a painless swelling on the neck, and Kepertis et al.[5] in their study found that 63.3% of their patients had a palpable cystic mass in the mid-neck region. In our study, we found a painless cystic midline mass in 48 (52.7%) patients, whereas 43 (47.3%) patients presented with a fistula in the midline neck area.

At diagnosis, the TGDC history and physical examination are the key points. Imaging modalities and FNAB can also be used. In patient's history, a painless cystic mass in the mid-neck region that moves with tongue protrusion, is the most common complaint. Less commonly, fistula with or without a draining sinus can be seen. USG is the most appropriate imaging modality since it is cheap, non-invasive, can be easily obtained, and a differential diagnosis with other neck masses can be made. For the lesions near the base of the tongue, MRI is a better choice.[12]

The Sistrunk procedure was performed in all our patients. In all cases, the corpus of the hyoid bone was excised with the tract of the cyst or fistula to decrease the recurrence rates. Bratu et al.[13] in their study found that the recurrence rates were 12% with the Sistrunk procedure. In a review of children operated for TGDC, the recurrence rate was 10.7%.[14] In the literature, recurrence rates ranged between 0% and 16%.[15] In our study, recurrence was seen in 14 (15.4%) of our patients, which was similar to literature.

There are some recurrence factors explained in the literature, and the most commonly blamed factor is preoperative infection. In a study of 270 patients, the authors concluded that in addition to preoperative infection, draining the cyst preoperatively was also a common reason for recurrence.[6] Bratu et al.[13] in their study concluded that the recurrence risk related to a preoperative infection was 27%. With regard to the TGDC recurrence rates, some other factors were found in the literature such as an inadequate cyst excision and young age (<2 years).[15] The average duration of recurrence was 4.93±7.16 years in our study. This may be a result of the age group of patients consulting to our otolaryngology clinic where younger patients may be seen and treated by pediatric surgeons. In our study, 10 out of 14 recurrence patients were patients with TF, and they had a history of recurrent infection. When compared to literature, we may also describe preoperative infection as the most common recurrence factor. We applied the Sistrunk procedure in all of the patients, and we had very low complication rates as well as recurrence rates as low as 3%. The Sistrunk procedure is a safe and well-tolerated procedure. Since recurrence is mostly related to preoperative infection, in patient selection, one should be aware of this and should plan the procedure after the infection is treated.

Similar to all surgical procedures, in the Sistrunk procedure, complications are possible. Superficial infection at the local wound site is the most common complication, and it is usually well tolerated.[16] Hematoma, undesired scar tissue, cervical edema, rupture of the cyst the during procedure, and recurrence are other possible complications. In our study, a total of 3 (3%) patients had a local wound site infection, 2 (2%) had hematoma, and 2 (2%) had undesired scar formation; none of them required surgical treatment.

CONCLUSION

Our study revealed that, in diagnosing TGDC, a physical examination and USG are the most useful techniques with low costs and easy access. The gold standard in the treatment of TGDC and TF is surgery. The Sistrunk procedure is the most commonly preferred procedure, with very low recurrence rates. The follow-up of patients after surgery is essential for diagnosing and treating recurrence.

Ethics Committee Approval

Kartal Dr. Lutfi Kirdar Training and Research Hospital Ethical Committee Approval Number: 2018/514/138/7.

Peer-review

Internally peer-reviewed.

Authorship Contributions

Concept: H.B.; Design: M.D.E.; Supervision: S.A.; Data collection &/or processing: H.B.; Analysis and/or interpretation: M.D.E.; Literature search: H.A.; Writing: M.D.E.; Critical review: M.D.E.

Conflict of Interest

None declared.

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Tiroglossal Duktus Kist ve Fistül Hastalarının Tanısal ve Cerrahi Değerlendirmesi: Yedi Yıllık Deneyimimiz

Amaç: Tiroglossal duktus kistleri (TGDK) çocuklarda orta hat boyun kitlelerinin başında gelmekle birlikte erişkin çağda da görülebilmektedir. Bu çalışmada amacımız TGDK tanısı almış hastaların demografik özelliklerini değerlendirmek ve TGDK'li hastaların tanı, tedavi planları ve takip detaylarını tartışmaktır.

Gereç ve Yöntem: Ocak 2010–Şubat 2017 tarihleri arasında kliniğimizde TGDK tanısı alan 91 hastaya ait veriler elektronik olarak toplandı. Veriler arasında demografik özellikler, tıbbi kayıtlar, ameliyat sonrası takip ve komplikasyonlar vardı. Tanı, fizik muayene, ultrasonografi (USG), bilgisayarlı tomografi (BT) ve manyetik rezonans görüntüleme (MRG) dahil olmak üzere görüntüleme yöntemleri ile yapıldı. Patoloji, çalışmaya dahil edilen tüm olgularda TGDK'yı doğruladı.

Bulgular: Doksan bir hastadan 49'u (%53) erkek, %46'sı kadındı. Hastaların yaş ortalaması 20.29 olarak bulundu. Tüm hastalara Sistrunk prosedürü uygulanmış olup, hastaların 14'ünde (%15) nüks saptanmıştır.

Sonuç: Her yaşta orta hat boyun kitlelerinin ayırıcı tanısında TGDK düşünülmelidir. Fizik muayene ve USG tanı koymada en kolay ve ucuz yöntemlerdir. TGDK tanısında cerrahi ana tedavi modalitesidir. Sistrunk prosedürü en düşük nüks oranına sahip olan altın standart cerrahi yöntemidir.

Anahtar Sözcükler: Fistül; kist; sistrunk operasyonu; tiroglossal duktus.