OLGU SUNUMU/CASE REPORT
J Turgut Ozal Med Cent 2014;21(2):148-50

Journal Of Turgut Ozal Medical Center
www.jtomc.org

A Rare Form of Thyroglossal Duct Cyst: Double Thyroglossal Cyst and a Review of the Literature

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Abstract
Thyroglossal cyst is the most common congenital mass in the neck. However, double thyroglossal cysts are very rare and our case is the sixth case that has ever been published in the literature. Cysts can be seen in any region between the base of the tongue and the supra-sternal region. These are painless cysts of soft consistency, which are movable with protrusion of the tongue and swallowing. Several imaging techniques are used to verify the diagnosis. Ultrasonography, for instance, is generally used to this end. However, in double cysts, we suggest that thyroid scintigraphy should be used in order to differentiate these cysts from aberrant thyroid tissues. In addition, treatment of double thyroglossal cysts is also successful through Sistrunk operations.
Key Words: Children; Double; Thyroglossal Cyst; Scintigraphy.

Tiroglossal Kistin Nadir Formu: Çift Tiroglossal Kist ve Literatür Değerlendirilmesi

Özet
Tiroglossal kist en sık görülen konjenital boyun kitlesidir. Bununla birlikte, çift tiroglossal kist son derece nadir olarak görülmektedir. Bizim sunmakta olduğumuz vaka günümüze kadar yayınlanmış altıncı vakadır. Tiroglossal kistler ise dil kökü ya da suprasternal bölge arasında herhangi bir bölgede görülebilir. Bu kistler ağrısız ve yumuşak kıvamlıdır ve dilin dışarı çıkartılması veya yutkunmakla hareketli olabilmektedir. Görüntüleme teknikleri daha çok sıklıkla tanıyı doğrulamak amacı ile kullanılır. Ultrason genellikle en sık kullanılan yöntemdir. Ancak çift tiroglossal kistlerde temel olarak aberan troid dokusundan ayırım için troid sintigrafisinin de yapılması gerektiğini düşünmekteyiz. Ayrıca çift tirolossal kistlerde klasik sistrunk operasyonu ile başarılı sonuçların elde edilebileceğini düşünmekteyiz.
Anahtar Kelimeler: Çocuk; Çift Tiroglossal Kist; Sintigrafi.

INTRODUCTION

Thyroglossal cysts (TGCs) are the most common congenital masses in the neck. They can be seen in any region between the base of tongue and supra-sternal region (1,2).

Thyroglossal duct, which is an epithelial tract formed during migration of thyroid gland in embryogenesis from base of mouth to its normal location in front of inferior part of the neck, normally disappears during embryonic life. It is estimated that these ducts do not disappear in 7% of the entire population. TGCs occur as a result of stimulation of epithelial remains of thyroglossal ducts that have not disappeared in embryonic life through upper respiratory tract infections (1-3). Double TGC is a very rare situation caused by the complete failure of obliteration of thyroglossal duct (3,4). This article presents a double TGC case, which is very rare in the literature.

CASE REPORT

A 6-year-old male patient presented to our clinic with the complaint of swellings in the neck. The first swelling had appeared a year ago and the second swelling had appeared three months ago. The swellings had been growing intermittently and there was no pain or erythema. On physical examination of the patient, there were two painless, mobile masses of soft consistency in the midline of the neck, the first in the infrahyoid region and the second, which was smaller than the first, in the suprahyoid region (Figure 1).

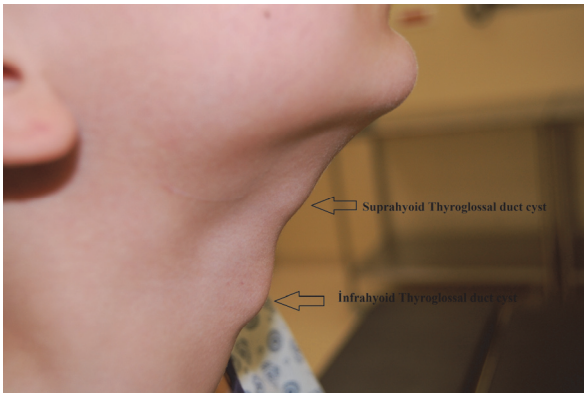

Figure 1. Double thyroglossal cyst in children

The masses were mobile with protrusion of the tongue. First, a neck ultrasonography (US) was performed. On

US, there was an intense cystic lesion measuring 14x5.4x19 mm in size in the midline of the neck and superior thyroid isthmus, and there was another intense cystic mass at the superior of this region measuring 15.9x8 mm in size close to the root of the tongue. Complete blood count, biochemical and thyroid function tests were normal. Thyroid scintigraphy was requested in order to eliminate the possibility of ectopic thyroid tissue, on which the thyroid gland was determined to be normal. The patient was operated and the Sistrunk procedure was performed. The perforation of the inferior cyst took place during the operation. The other cyst was removed totally together with the hyoid bone corpus (Figure 2). The patient was discharged on the 3rd postoperative day uneventfully. We did not encounter recurrence during the one-year follow-up. Histopathological examination showed that both cysts were lined with a squamous epithelium and columnar epithelium.

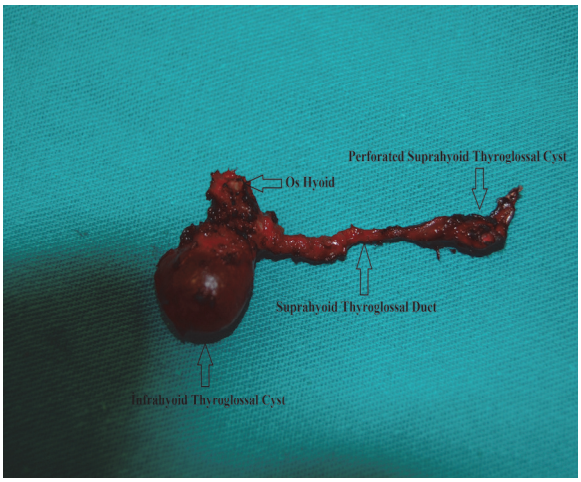

Figure 2. Macroscopic aspect of double thyroglossal cyst

DISCUSSION

Abnormalities of the thyroglossal duct develop from partial or total lack of obliteration of the thyroglossal duct. Accordingly, TGC or versions of TGC (single or double fistula tract, single or double cyst TGC) may develop. Thyroglossal cysts are the most common congenital masses in the neck. In children, it comprises more than 75% of the midline or infrahyoid masses. However, double TGC is very rare. Our literature search showed five previously reported cases, thus marking our case the sixth double TGC case (Table 1) (2,3,5).

In spite of the fact that these anomalies are seen at the same rates in both genders, they can be seen anytime during the course of life; yet they are noteworthy in children, particularly in the first five years of childhood (3). TGCs can develop at any location on the persistent thyroglossal duct. In 60% of the cases, they are located between the hyoid bone and the thyroid tissue; in 24% of the cases, they are located in the supra hyoid region; in 13%, they are suprasternally located, and in 1% of the cases, they are intralingually located (2).

Accurate diagnosis is commonly made with history and physical examination. TGCs are masses in the neck midline that are mobile with swallowing and protrusion of the tongue. The cysts are generally painless, slightly mobile, asymptomatic soft masses. Thyroglossal cysts can sometimes be infected and fistula may develop. They can sometimes cause dysphagia and hoarseness. Imaging techniques are commonly used to confirm the diagnosis. Ultrasonography is commonly used for the diagnosis (1-4,6). For an accurate diagnosis, a differential diagnosis should be made with dermoid cyst, normal thyroid tissue and thyroid malignancies, aberrant thyroid tissue, branchial cleft cyst, lipoma, lymphadenopathy, hemangioma, lymphangioma, and teratoma[3]. Ultrasonography, computerized tomography, magnetic resonance and thyroid scintigraphy are used for the differential diagnosis (1,2). Arguments about the evaluation of preoperative TGC with thyroid scintigraphy still continue. However, its use for distinguishing the pathologies of the thyroid tissue and ectopic thyroid tissue are recommended (7). We utilized US and thyroid scintigraphy for the preoperative diagnosis of our patient. We used thyroid scintigraphy for thyroid malignancy and to distinguish ectopic thyroid tissue that can possibly be found be within the TGC. We suggest that scintigraphic evaluation is important in distinguishing malignancy and ectopic thyroid tissue, particularly when there is suspicion of double TGC.

Table 1. Double TGCs in the literature

Author	Gender	Age	Presentation
Pueyo et al[10] (2008)	Male	7 Years	The hyoid region and in the thyroid gland
Khadivi et al[5] (2010)	Female	14 years	Infrahyoid
Bora et al[3] (2011)	Female	9 Years	Unknown
Yorgancılar et al[4] (2012)	Unknown	Unknown	The hyoid region and the tongue base
Sarmento et al[2] (2013)	Female	30 years	Floor of the Mouth and Sublingual Gland
Our Patient	Male	6 Years	Infrahyoid and suprahyoid

Surgical excision is commonly used as the mode of treatment. Recently, the recommended surgical option has been the Sistrunk operation in which the removal of the hyoid bone corpus is additionally performed. A recurrence rate of 1.5-5% has been defined after the operation, and a recurrence rate of 20-38% has been determined in patients with hyoid bone untouched (3,8). Recently, as an alternative to surgical treatment, injection of sclerosing substance with ethanol into the cyst has been discussed (9). Surgeons used Sistrunk operation for double TGCs. None of these patients reported recurrence (2-5,10). In our case, we, too,

employed the Sistrunk operation. We think that Sistrunk operation is satisfactory for the treatment of double TGCs.

Histopathologically, the microscopic findings of the cyst vary according to the location. Cysts and fistulas on the hyoid bone were lined with squamous epithelium, but below the hyoid bone cysts and fistulas were lined with columnar epithelial (3). However histopatological examination of double TGC cases showed that both cysts and the connecting tract were lined with a squamous epithelium with cylindrical and ciliated epithelial areas of respiratory type (2,10). Microscopic examination of our case was in line with literature.

Malignant degeneration can be seen in 1% of TGCs. The diagnosis of malignant degeneration can be made with postoperative histopathological examination. Eighty-five percent of malignancies are papillary carcinomas. More rarely, mixed papillary carcinoma, squamous cell carcinoma, adenocarcinoma and anaplastic carcinoma have likewise been observed (8,9). Malignancy was not detected in our case and other double TGCs.

Consequently, double TGCs are very rare. Commonly, the preoperative diagnosis is made with physical examination and US. However, we suggest that thyroid scintigraphy is important in order to make the differentiation between thyroid pathologies and double cyst TGC. Classic Sistrunk operation is satisfactory for the treatment of double TGCs.

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Received/Başvuru: 25.08.2013, Accepted/Kabul: 09.10.2013

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For citing/Atıf için

Yildiz T, Ilce HT, Kucuk A, Ilce Z. A rare form of throglossal duct cyst: double thyroglossal cyst and review of the literature. J Turgut Ozal Med Cent 2014;21:148-50 DOI: 10.7247/jtomc.2013.1213