A Very Rare Cause of Subglottic Stenosis: Non-Malignant Intratracheal Thyroid Tissue

Subglottik Stenozun Çok Nadir Bir Nedeni: İntratrakeal Malign Olmayan Tiroid Dokusu

İntratrakeal Malign Olmayan Tiroid Dokusu / Non-Malignant Intratracheal Thyroid Tissue

Ümit Aydoğmuş[1], Figen Türk[1], Gökhan Yuncu[2]
[1]Göğüs Cerrahisi, Pamukkale Üniversitesi Tıp Fakültesi, Denizli,
[2]Göğüs Cerrahisi Kliniği, Liv Hospital, İstanbul, Türkiye

Özet

Burada multinodüler guatr nedeniyle 14 yıl önce opere olmuş bir hastada nüks-le birlikte benign tiroid dokusunca invazyona bağlı subglottik stenozlu bir olgu sunulmuştur. 14 yıl önce bilateral subtotal tiroidektomi geçirmiş hastanın patolojisi multinodüler tiroid dokusu olarak raporlanmıştı. Ani solunum sıkıntısı gelişmesi üzerine acil servise getirilen hasta merkezimize yönlendirmişti. Servikal tomografisinde tiroid bezi seviyesinde trakeaya sol yan duvardan invazyon göstererek trakea çapını %80 oranında daraltan bilateral tiroid dokusu izlendi. Ciddi solunum sıkıntısı nedeniyle acil trakeostomiye alınan hastanın trakeası krikoid kartilajın 2,5 cm kadar altından vertikal insize edildi. Sol kord vokalden sınırı ayrıt edilmeyen 2 cm boyunca devam eden endotrakeal lezyon izlendi, hem bu lezyondan hem de trakea çevresindeki dokudan biyopsiler alındı. Subglottik alandan distale uzanan Montgomery T tüp yerleştirildi. Patolojisinde normal tiroid dokusu ile uyumlu histopatolojik bulgular izlendi. Tiroid kanserine bağlı trakea invazyonu sık olmamakla birlikte oluşabilmektedir. Biz benign nedenli tiroid cerrahisi sonrası intratrakeal nüks gelişmiş bir olgu ve onun acil tedavisi sunduk

Anahtar Kelimeler

Subglottik Stenoz; İntratrakeal Tiroid; Guatr Ameliyatı Komplikasyonu

Abstract

We present a case of subglottic stenosis associated with benign thyroid tissue involvement due to relapse of multinodular goiter despite surgery 14 years ago. The patient had undergone bilateral subtotal thyroidectomy 14 years ago and the pathology report had been multinodular thyroid tissue at the time. The patient recently presented to an emergency service due to sudden development of respiratory distress and was then directed to our center. Cervical tomography showed bilateral thyroid tissue that narrowed the tracheal diameter by 80% by invading the trachea from the left wall at the level of the thyroid gland. The patient required urgent tracheostomy due to serious respiratory trouble. The trachea was incised vertically about 2.5 cm below the cricoid cartilage. A 2 cm endotracheal lesion with margins that could not be distinguished from the left vocal cord was observed and biopsies were taken from both this lesion and the tissue surrounding the trachea. A Montgomery T-tube extending from the subglottic area to the distal section was placed. Pathology evaluation revealed histopathological findings that matched normal thyroid tissue. Although infrequent, tracheal invasion associated with a thyroid cancer is known to occur. We present a case with postoperative intratracheal relapse due to a benign cause and the emergency treatment.

Keywords

Subglottic Stenosis; Intratracheal Thyroid Tissue; Complication of a Goiter Operation

DOI: 10.4328/JCAM.3370 Received: 07.03.2015 Accepted: 31.03.2015 Printed: 01.08.2014 J Clin Anal Med 2014;5(suppl 4): 521-3
Corresponding Author: Ümit Aydoğmuş, Göğüs Cerrahisi A.D. Pamukkale Üniversitesi Tıp Fakültesi, Denizli, Türkiye.
GSM: +905512614232 F.: +90 2582961765 E-Mail: mdaydogmus@yahoo.com

Introduction

The causes of benign tracheal stenosis include intubation, tracheostomy, chemical or thermal damage and trauma, and less frequently inflammatory or connective tissue diseases, sarcoidosis and idiopathic subglottic stenosis [1]. Tracheal injury during a thyroid surgery is very rare [2, 3]. Minor iatrogenic rupture can go unnoticed and reappear in very late periods as a relapse of the disease or subglottic stenosis. We present a patient who had undergone bilateral subtotal thyroidectomy due to multinodular goiter and developed a subglottic stenosis associated with intratracheal thyroid tissue 14 years after the surgery for the first time in the literature.

Case Report

A 37-year-old female patient underwent bilateral subtotal thyriodectomy 14 years ago and the pathology report was multinodular thyroid tissue. The patient had been using asthma treatment for the last three years. The respiratory problems particularly the shortness of breath and stridor had increased recently. She presented to the emergency service of an external health care facility because of acute respiratory distress. The indirect laryngoscopy by the otolaryngologist revealed a tracheal mass under the vocal cords and she was referred to our center. Her stridor could be heard without auscultation during her physical examination. A soft tissue formation sized 15x12 mm was seen on her cervical computed tomography. The mass was localized on the left subglottic area and extended towards the lumen, causing about 80% stenosis in the lumen of the larynx and trachea. There was heterogeneous contrast enhancement involvement after an intravenous contrast medium injection and the margins could not be distinguished clearly from the left thyroid lobe (Figure 1). The margins also could not be



Figure 1. Computed cervical tomography coronal plane

distinguished from the left vocal cord on the superior aspect (Figure 2). Both thyroid lobes seemed to be increased in size and had a heterogeneous appearance. Additionally, heterogeneous iso-hypoechoic solid nodular lesions were seen in both thyroid lobes; the largest was 19 mm in size and located on the posterior aspect of the middle part of the left thyroid lobe on cervical computed tomography (Figure 3).

Urgent tracheostomy was required due to serious respiratory distress. The trachea was incised vertically about 2.5 cm under cricoid cartilage. There was a 2 cm endotracheal lesion whose



Figure 2. Computed cervical tomography sagittal plane



Figure 3. Computed cervical tomography transverse plane

borders with the left vocal cord could not be distinguished. The left side of this area was invaded by a thyroid tissue. Many biopsies were taken from both this lesion and the tissue surrounding the trachea. Intratracheal tissue was removed as much as possible by electrocautery and sharp dissection. A Montgomery T-tube was placed extending from the subglottic area to the distal section (Figure 4). The respiratory difficulty was relieved



Figure 4. Postoperative lateral graphy

after the surgery. The pathology result was reported as thy-

roid tissue. Thyroid I-131 scintigraphy was performed after the patient's breathing became stable and revealed a hyperplastic normoactive thyroid gland that showed invasion into the trachea.

The patient is being monitored and is now in the 7th postoperative month with no problems.

Informed Consent: Written informed consent was obtained from patient who participated in this case.

Discussion

The causes of benign stenoses of the subglottic tracheal area involve post-intubation and tracheostomy, and rarely trauma and idiopathic situations. Iatrogenic tracheal ruptures are extraordinarily rare and have most commonly been described after manipulations such as intubation and bronchoscopy [2]. Tracheal ruptures occurring during surgery on the thyroid are usually noticed during the procedure [3]. Gosnel et al. [3] have reported tracheal rupture during surgery in only one case (0.06%) out of 11917 thyroid operations and primary repair was performed during the procedure. A few cases that resulted in tracheal necrosis and rupture in the late postoperative period due to excessive cautery or prolonged intubation have been reported [2, 4].

Thyroid tumors are known to cause transmural invasion of the trachea [5]. The cause is trachea obstruction in nearly a half of fatal papillary thyroid carcinomas [5]. We are the first to report involvement of the trachea by benign thyroid tissue. Moreover, the patient presented with acute respiratory distress 14 years after her first operation. The biopsies taken from both inside the trachea and the tissue outside the trachea revealed benign thyroid tissue. In view of these findings, we considered a relapse that also involved an endotracheal component occurring many years later due to an unnoticed small tracheal rupture during the initial thyroid surgery.

Benign tumors of the trachea are rare and both short- and long-term results of endoscopic treatment are satisfactory [6]. Surgical treatment is recommended when the tumor base is not entierely visible, as in the present case [7]. Subglottic stenoses are complex problems [5]. The location and size of the lesions and the complications due to obstruction are important factors in the choice of the treatment method [6]. Patients with benign tracheal tumors that usually grow slowly may have received treatment for asthma for a long time [6]. Unfortunately, our case was not diagnosed until emergency intervention was required for acute respiratory distress. Endoscopic treatment modalities were not used in our patient as the base of the tumor was not entirely visible and an emergency airway had to be provided. The patient underwent urgent tracheostomy. A T-tube was preferred instead of standat tracheostomy canule for protecting the vocal cords functions. Major surgery was not performed as the left vocal cord was seen to have been involved by the thyroid tissue during the procedure. Using a T-tube instead of a tracheostomy has the advantages of enabling speech, normal humidification of the trachea, causing less mucosal damage and acting as a stent on the tube's both proximal and distal aspect. It also restores airways temporarily or permanently in situations where there is a non-removable lesion or it is impossible to perform reconstruction [8].

We have presented a case with benign intratracheal thyroid tissue that caused subglottic stenosis as a very late complication of thyroidectomy 14 years ago for the first time in the literature. Benign tumors of the trachea can be treated by endoscopy but surgery may sometimes be needed. Tracheostomy is needed very rarely. Using a T-tube may be an option if it is not possible to remove the lesion.

Competing interests

The authors declare that they have no competing interests.

References

1. Perotin JM, Jeanfaivre T, Thibout Y, Jouneau S, Lena H, Dutau H, et al. Endoscopic management of idiopathic tracheal stenosis. Ann Thorac Surg 2011;92(1):297-301.
2. Mazeh H, Suwanabol PA, Schneider DF, Sippel RS. Late manifestation of tracheal rupture after thyroidectomy: case report and literature review. Endocr Pract 2012;18(4):73-6.
3. Gosnell J E, Campbell P, Sidhu S, Sywak M, Reeve TS, Delbridge W. Inadvertent tracheal perforation during thyroidectomy. Br J Surg 2006;93(1):55-6.
4. Chauhan A, Ganguly M, Saidha N, Gulia P. Tracheal necrosis with surgical emphysema following thyroidectomy. J Postgrad Med 2009;55(3):193-5.
5. Yang CC, Lee CH, Wang LS, Huang BS, Hsu WH, Huang MH. Resectional treatment for thyroid cancer with tracheal invasion: a long-term follow-up study. Arch Surg 2000;135(6):704-7.
6. Ceylan KC, Akpınar D, Usluer O, Ünsal Ş, Polat H, Aydoğdu Dinç Z. Leiomyoma of the Trachea: Case report. Eurasian J Pulmonol 2011;3(2):125-7.
7. Dalar L. Trakeanın benign darlıklarında girişimsel bronkoskopik yöntemler. In Bedirhan MA editor, Trakea. Istanbul: Probiz Ltd Şti; 2012. p.129-57.
8. Grillo HC. Development of Tracheal Surgery: A Historical Review. Part 1: Techniques of Tracheal Surgery. Ann Thorac Surg 2003;75(2):610-9.

How to cite this article:
Aydoğmuş Ü, Türk F, Yuncu G. A Very Rare Cause of Subglottic Stenosis: Non-Malignant Intratracheal Thyroid Tissue. J Clin Anal Med 2014;5(suppl 4): 521-3.