Bezmialem Science 2017; 5: 138-41
DOI: 10.14235/bs.2016.1074

An Adult Tracheocele with No Predisposing Factor

Predispozan Faktör Olmaksızın Erişkin Trakeosel

Burak ERTAŞ[1], Tuncay ULUĞ[2], Kubilay AYDIN[3]

[1]Department of Otorhinolaryngology, Acıbadem University School of Medicine, İstanbul, Turkey
[2]Clinic of Otorhinolaryngology, Uluğ Klinik, İstanbul, Turkey
[3]Department of Radiology, İstanbul Üniversitesi İstanbul School of Medicine, İstanbul, Turkey

ABSTRACT

Tracheoceles are rare, and usually, they have been described as incidental findings while evaluating patients for other problems. Our patient complained of a mass located in the right supraclavicular region that got larger on coughing and straining; otherwise, he was asymptomatic. His history did not reveal any predisposing factors. Computed tomography showed an air-filled 3×2.5×2 cm mass at the level of the T2–4 vertebrae. Surgical exploration showed an air-filled mass located between the common carotid artery and trachea, communicating with the tracheal lumen via a narrow tract attached to the posterior wall of the trachea. The mass was completely resected, and the defect in the posterior wall was repaired. A literature search revealed only one tracheocele case without any predisposing factors, and our case is a new one. It is different from other tracheocele reports considering the origin side, type, and level of the lesion.

Keywords: Tracheocele, diverticulum, neck, swelling

ÖZ

Trakeoseller nadir görülür ve genellikle hastalar diğer problemleri için değerlendirilirken tesadüfen saptanır. Hastamız öksürük ve gerilme ile genişleyen asemptomatik sağ supraklavikuler kitle şikayeti ile başvurdu. Hikayesinde predispozan bir faktör görülmedi. Bilgisayarlı tomografisinde T2-4 vertebralar seviyesinde hava ile dolu 3x2,5x2 cm ölçülerinde kitle görüldü. Cerrahi girişimde kitlenin ana karotis arter ile trakea arasında olduğu ve dar bir bağlantı ile trakea posterior duvarı ile bağlantısı olduğu görüldü. Kitle tam olarak çıkarıldı ve posterior duvarda defekt tamir edildi. Literatürde bizim olgumuz gibi predispozan faktör olmaksızın gelişen sadece bir olgu bildirilmiştir. Bizim vakamız literatürdeki diğer vakadan seviyesi, kaynaklandığı yer ve tipi farklıdır.

Anahtar Kelimeler: Trakeosel, divertikulum, boyun, şişlik

Introduction

Tracheoceles have rarely been described. Some have been described as incidental findings while evaluating patients for other problems such as carcinoma of the larynx, in association with trachiectasis, as a complication of tracheostomy, or following the development of pneumomediastinum after a difficult endotracheal intubation (1-4). Mathur et al. (5) reported a patient with a large tracheocele presenting in the neck with no predisposing factor, which was similar to that seen in our patient (5). The main symptoms of tracheoceles are cough and purulent expectoration. The presence of symptoms is an indication for surgery. The suggested surgical approach is resecting the mass and tract communicating with the trachea and suturing the tracheal defect.

Case Report

A 43-year-old male presented with right-sided swelling in the neck. There was no stridor, hoarseness, or dysphagia. He had no predisposing factor for a tracheocele, such as trumpet playing, glass blowing, intubation history, tracheostomy, pulmonary disease, or trauma to the neck. The patient was informed about the surgery, and written consent was obtained. His physical examination revealed a swelling in the right supraclavicular region, which increased in size during the Valsalva maneuver. Laryngeal endoscopy revealed normal findings. Computed tomography (CT) of the neck revealed a unilocu-

Cite this article as: Ertaş B, Uluğ T, Aydın K. An Adult Tracheocele with No Predisposing Factor. Bezmialem Sicemce 2017; 5: 138-41.

Address for Correspondence/Yazışma Adresi: Burak ERTAŞ; Acıbadem Üniversitesi Tıp Fakültesi, Kulak, Burun, Boğaz Anabilim Dalı, İstanbul, Türkiye E-mail: drbertas@yahoo.com

©Copyright 2017 by Bezmialem Vakif University - Available online at www.bezmialemscience.org
©Telif Hakkı 2017 Bezmialem Vakif Üniversitesi - Makale metnine www.bezmialemscience.org web sayfasından ulaşılabilir.

Received / Geliş Tarihi : 05.05.2016
Accepted / Kabul Tarihi: 18.06.2016
Available Online Date / Çevrim İçi Yayın Tarihi : 06.07.2017

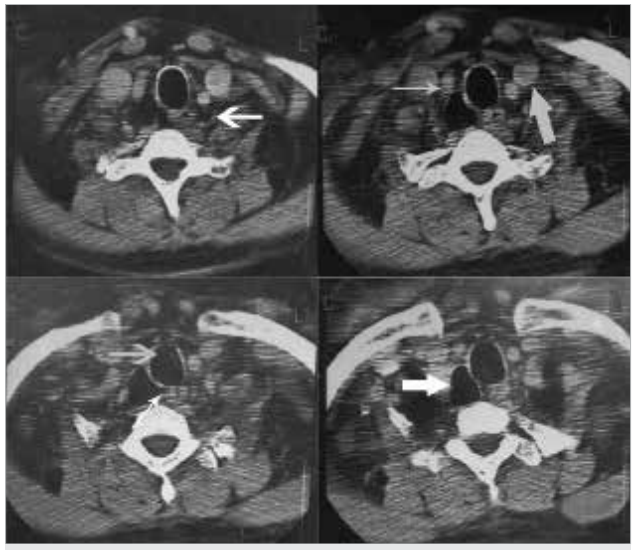


Figure 1. Preoperative CT image of our patient showing communication between the trachea and the tracheocele (small white arrow), esophagus (medium white arrow), tracheocele sac (big white arrow), right common carotid artery (small grey arrow), trachea (medium grey arrow), and internal jugular vein (big grey arrow)

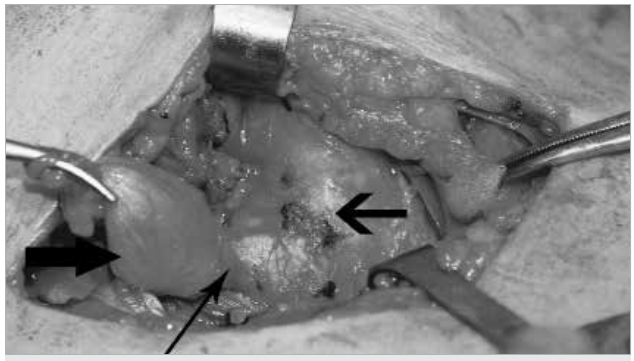


Figure 2. Intraoperative image showing communication between the trachea and the tracheocele (small arrow), wide shallow trachea (medium arrow), and sac (big arrow)

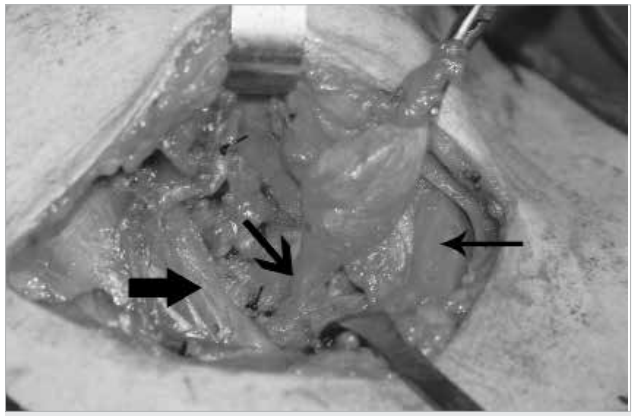


Figure 3. Intraoperative image showing the strap muscles (small arrow), communication between the trachea and tracheocele (medium arrow), and sternocleidomastoid muscle (big arrow)

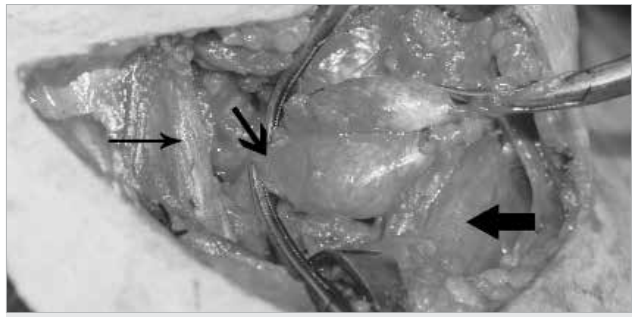


Figure 4. Intraoperative image showing the strap muscles (small arrow), communication between the trachea and the tracheocele (medium arrow), and sternocleidomastoid muscle (big arrow)

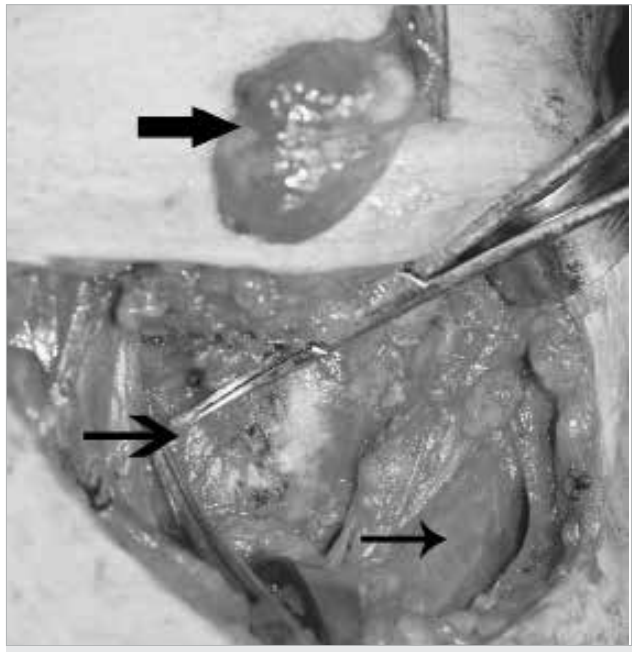


Figure 5. Intraoperative image showing the sternocleidomastoid muscle (small arrow), communication between the trachea and the tracheocele (medium arrow), and sac (big arrow)

lar air-filled cavity measuring 3×2.5×2 cm at the level of the T2–4 vertebrae on the right posterolateral side of the trachea, which displaced the trachea to the left. The CT images showed continuity between the trachea and the lesion. No communication with the laryngeal ventricle was observed (Figure 1). Posteroanterior X-rays were taken with and without the Valsalva maneuver, and a unilocular cavity corresponding to the one seen on performing CT was found.

Preoperative laboratory examination results were normal. The lesion was approached via a horizontal supraclavicular incision, and the region between the trachea and the right common carotid artery was explored. The air-filled mass was oval, transilluminating, and did not contain any fluid. During surgery, a communication between the mass and the fibroelastic portion of the posterolateral wall of the eight and ninth tracheal cartilages was found (Figures 2–4). The mass

laterally displaced the common carotid artery and internal jugular vein. The trachea was widened and compressed by the lesion (Figure 2). The mass was dissected from the soft tissues and the wall of the trachea and was resected in one piece, including the tract communicating with the trachea (Figure 5). The tracheal defect was closed with 3.0 vicryl sutures (Figure 5). No communication with other parts of the trachea or larynx was seen. Tracheostomy was not performed. The postoperative period was uneventful, and the patient was followed for 2 years without any recurrence.

Discussion

Unlike laryngoceles, tracheoceles are extremely rare, although their true incidence is unknown (1, 2, 5). A literature search revealed only one tracheocele case without any predisposing factor (5). Typically, the tracheocele in the reported case originated in the right posterolateral side of the trachea (5), although Andersen *et al*. reported a diverticulum that occurred on the left side (2, 5). In our patient, whose condition was similar to the patient's condition reported by Mathur *et al*., there was no stridor, hoarseness, or dysphagia, and both patients presented with gradually progressive right-sided swelling of the neck that became larger with the Valsalva maneuver. In both patients, the laryngeal examination and laboratory findings were normal (5). Alt et al. and Nerurkar *et al*. reported two patients having a tracheocele and complaining of progressive dysphonia. An endoscopic examination of the larynx in both patients revealed right true vocal cord paralysis (6).

In our case, the radiological investigation and operative findings confirmed that the mass was an air-filled cavity that was firmly attached to the right posterolateral trachea near its lower end. This finding is consistent with that observed in the literature. Furthermore, the mass was unilocular, similar to that observed in reported cases (5).

The differential diagnosis of such swellings includes a laryngocele, goiter, a cystic hygroma, a large Zenker's diverticulum, and phlebectasia of the internal jugular vein. The main features distinguishing a tracheocele are the air-filled cavity and increased size of the swelling with the Valsalva maneuver (7).

Miller reported that the trachealis muscle consists of transverse bands that unite with the cornua of C-shaped tracheal cartilage rings (8). These bands are separated by connective tissue. The development of a diverticulum or tracheocele in these relatively weak areas is possible, particularly if there is an infection in the mucous membrane and chronic cough with increased intrabronchial pressure (4, 8). It is not easy to tell whether tracheoceles are congenital or acquired, although it is generally accepted that tracheoceles with wide openings are acquired lesions, whereas those with narrow openings are congenital (9). The opening in our patient was narrow, and the trachea at this level was wide and shallow. The lesion was suspected of being congenital as the opening was narrow, the patient was relatively young, the symptoms had been present for a long time, and there was no predisposing factor.

In contrast to some reports, the origin of the lesion was obvious. There was an obvious communication between the mass and the trachea, and no communication was seen with the laryngeal ventricle.

In our patient, the sac was located between the eighth and ninth tracheal cartilages, while in the patient of Mathur et al. (5) it was between the ninth and tenth cartilages. We dissected the sac from the soft tissues and wall of the trachea and resected it in one piece, including the communication with the trachea. Unlike Mathur *et al*., we did not perform tracheostomy. Berlucchi et al. (10) performed brushing of the tracheal diverticulum associated with the application of fibrin glue into the pouch using an endoscopic approach in a pediatric patient who complained of recurrent pneumonitis episodes. This approach seems to be appropriate for small masses of the tracheocele, on which surfaces can be accessed for brushing and applying the fibrin glue. Our patient was not appropriate for such an approach because of the measurements of the sac (10).

Conclusion

We demonstrated the relationship between the rare case of the tracheocele and trachea by intraoperative images. We think that the described relationship and accumulation of these cases in the literature will shed light on planning treatment in these patients in the future.

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - T.U.; Design - B.E.; Supervision - K.A.; Funding - B.E.; Materials - T.U.; Data Collection and/or Processing - B.E.; Analysis and/or Interpretation - K.A.; Literature Review - B.E.; Writing - T.U.; Critical Review - B.E.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

Hasta Onamı: Yazılı hasta onamı bu çalışmaya katılan hastalardan alınmıştır.

Hakem Değerlendirmesi: Dış bağımsız.

Yazar Katkıları: Fikir - T.U.; Tasarım - B.E.; Denetleme - K.A.; Kaynaklar - B.E.; Malzemeler - T.U.; Veri Toplanması ve/veya İşlemesi - B.E.; Analiz ve/veya Yorum - K.A.; Literatür Taraması - B.E.; Yazıyı Yazan - T.U.; Eleştirel İnceleme - B.E.

Çıkar Çatışması: Yazarlar çıkar çatışması bildirmemişlerdir.

Finansal Destek: Yazarlar bu çalışma için finansal destek almadığını belirtmiştir.

References

1. Gronner AT, Trevino RJ. Tracheocele. Br J Radiol 1971; 44: 979-981. [CrossRef]
2. Andersen HA, Clagett OT, Good CA. Trachiectasis with tracheocele: report of case in which a tracheocele was resected. Dis Chest 1953; 24: 553-7. [CrossRef]
3. Coret-Moya M, Moliner S, Valía JC, de Andrés J. Iatrogenic tracheocele after percutaneous tracheostomy. Rev Esp Anestesiol Reanim. 2013 Jan 30. doi:pii: S0034-9356(12)00472-0.
4. Möller GM, Berge EJFM, Stassen CM. Tracheocele: a rare cause of difficult endotracheal intubation and subsequent pneumomediastinum. Eur Respir J 1994; 7: 1376-7.
5. Mathur NN, DNB, Sardana P, Singh VP, Bais AS. Adult tracheocele with large cervical presentation. J Laryngol Otol 1999; 113: 364-5. [CrossRef]
6. Nerurkar N, Patil P, Tampi S, Yadav K, Jain S. Tracheocele--a case report. Laryngoscope 2011; 121: 1735-7. [CrossRef]
7. Lubianca-Neto JF, Mauri M, Prati C. Internal jugular phlebectasia in children. Am J Otorhinolaryngol 1999; 20: 415-8. [CrossRef]
8. Stocks RMS, Milburn M, Thompson J. Unusual neck masses secondary to jugular venous abnormalities: Case report and discussion. Amer Surg 1997; 63: 305-9.
9. Stibbe EP. True congenital diverticulum of the trachea in a subject also showing right aortic arch. J Anat 1929; 64: 62-6.
10. Berlucchi M, Pedruzzi B, Padoan R, Nassif N, Stefini S. Endoscopic treatment of tracheocele in pediatric patients. Am J Otolaryngol 2010; 31: 272-5.[CrossRef]