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# Tracheocele; a case of rare clinical entity

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## ÖZET

**Trakeosel; nadir bir klinik antite**

*Trakeosel trakeal mukoz membranın herniasyonudur ve literatürde çok nadir olarak rapor edilmiştir. Konjenital defekt veya sonradan kazanılmış bir lezyon şeklinde olabilir. Travmalar, yüksek basınçlı yaralanmalar, uzun sürede sonlandırılmış trakeostomiler, obstrüktif trakea hastalıkları, tekrarlayan infeksiyonların trakeanın mukus glandlarının kanallarını tıkmaması ve dilatasyonuna sebep olması gibi faktörler etyolojide rol oynar. En iyi tanı yöntemleri endoskopik muayene ve bilgisayarlı toraks tomografisidir. Kazanılmış trakeoselde tedavi yaklaşımı öncelikle konservatif olmalı ancak semptomatik olgularda cerrahi tercih edilmelidir.*

**Anahtar Kelimeler:** Trakea hastalıkları, spiral bilgisayarlı tomografi, bronkoskopi.

## SUMMARY

**Tracheocele; a case of rare clinical entity**

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*Tracheocele is a herniation of the tracheal mucous membrane and it is rarely reported in the literature. It may be a congenital defect or an acquired lesion. Traumas, high pressure injuries, long lasting tracheostomy, obstructive tracheal diseases,*

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recurrent infections of the mucous glands of the trachea with subsequent ductal obstruction and dilatation may play a role at the etiology. The best diagnostic procedures are endoscopic examination and computed tomography. The management of acquired tracheocele is primarily conservative, but surgical intervention may be indicated for symptomatic cases.

**Key Words:** Tracheal disease, spiral computed tomography, bronchoscopy.

Tracheocele has been rarely described in the literature, most often reported as an incidental finding during the evaluation of the patient with other complaints. It is an out-pouching of trachea lined by respiratory epithelium. Right side is involved more frequently. It results from a congenital or an acquired weakness of the tracheal wall which generally follows an obstructive tracheal disease and the diagnosis is often based on computed tomographic (CT) scan findings (1,2).

Absence of chronic lower respiratory tract infection suggests evidence of a congenital etiology. Trauma, infection and high pressure injury are the proposed etiology in acquired ones and can cause a variety of chronic and recurrent aerodigestive tract symptoms. Management is primarily conservative in asymptomatic cases (3,4).

We present the chest computed tomographic scan and fiberoptic bronchoscopy findings of an incidentally found asymptomatic case of tracheocele.

### CASE REPORT

A 66-year-old male admitted to our hospital with intermittent left chest pain aggravated by exercise. The patient was a farmer and he was a heavy smoker for 50 years. He had been hospitalized for pleural effusion with an unknown etiology and received a 35 days medical treatment 45 years ago. The auscultation of the lungs and physical examination for other organs were normal except a post nasal drip. Electrocardiography and Bruce test excluded coronary artery disease. Haematological and biochemical tests were all within normal range. The chest X-ray demonstrated pleural calcifications on the left side (Figure 1). Water's X-ray demonstrated unilateral maxillary sinusitis.



**Figure 1.** Posteroanterior chest X-ray showing pleural calcification at the left hemithorax.

Sputum microscopy and culture was negative for non-specific infection or tuberculosis. Chest CT scan showed an air filled cyst at the right posterolateral wall of trachea and pleural thickening and calcifications on the left lower zone (Figure 2). An  $^{18}\text{F}$ -FDG-PET/CT examination excluded malignancy. There was no obstructive or restrictive impairment in the lung function tests.

A fiberoptic bronchoscopy was performed under local anesthesia and small multiple orifices were detected at the membranous part of trachea. They were positioned 8 cm distal to vocal cords and arranged like a railway pattern, widest one being 2 mm in diameter. A bulging on the right tracheal wall was also noted (Figure 3).

Surgical management was not planned and chest pain disappeared in a week after receiving a nonsteroidal anti-inflammatory drug.

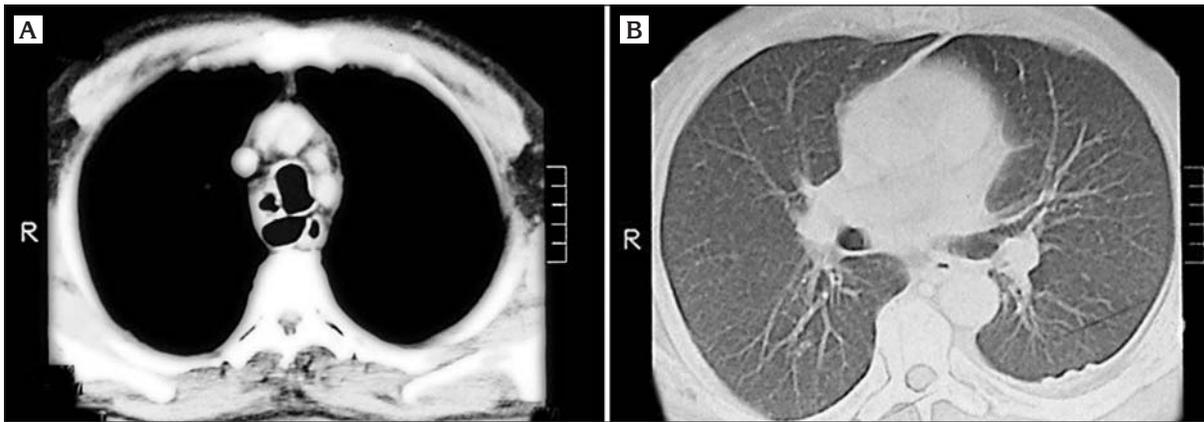


Figure 2. A. Chest CT scan showing the tracheocele at the right posterolateral wall of trachea. B. Chest CT scan showing pleural thickening and calcifications on the left lower zone.

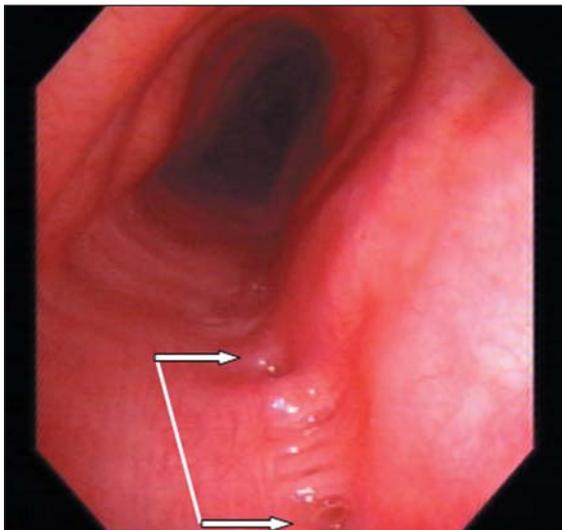


Figure 3. FOB image: Multiple orifices at membranous trachea arranged like a railway pattern (between the arrows). Also note the bulging of the right tracheal wall.

### DISCUSSION

Air filled cysts originating from trachea are extremely rare and their exact nature, whether it is a tracheocele, diverticulum or other form of air-filled cyst, remains a matter of debate. Their frequency is probably underestimated because most cause few symptoms and are well tolerated. In a recent radiological study their prevalence was found to be 2% in patients undergoing CT scan (5).

Tracheocele is a herniation of the tracheal mucous membrane (4). It is first described by Rokitsky in 1846 and later MacKinnon described

10 cases as an incidental finding of the autopsy procedure (6,7). It may be congenital or acquired. When congenital it is generally associated with small openings (8). Acquired tracheocele normally has a wide tracheal communication and trauma, high pressure injury, long lasting tracheostomy may be the etiologic factor. Beside these factors recurrent infections of the mucous glands of the trachea with subsequent ductal obstruction and dilation may play an accessory role (1,4). Our patient had no history of trauma and he was hospitalized for pleural effusion years ago. The orifices are multiple and small and the etiology was probably due to the past infection.

The most prominent sign of tracheocele is chronic secretive cough (4). It is thought to be caused by deposition of bronchial mucous within the defect. The other symptoms are bloody sputum and dysphagia (5). Although the patient is a heavy smoker and has postnasal dripping he doesn't suffer from cough. Besides, the small size of the orifices may limit collection of mucous in the cavity. In our patient chest pain was the unique symptom which was treated with a nonsteroidal anti-inflammatory drug.

Diagnostic studies include chest X-ray, CT, and bronchoscopy (9,10). Barium swallow may be performed to show the relationship with oesophagus which is mostly normal (2). On a CT analysis Han MC described that 98% of the paratracheal air cysts were located at the right posterolateral aspect of the trachea and 88% was at

T1-T2 vertebral levels (11). At fiberoptic bronchoscopy the orifice in the tracheal wall appear as tiny, well-circumscribed hole (10). Our patient's tracheocele was located at right posterolateral tracheal wall which is consistent with literature.

The management of tracheocele is primarily conservative, but surgical intervention may be indicated for patients with refractory symptoms (1). Complete dissection of the tracheocele and closure of the small communications by a running suture have been performed in symptomatic cases (3,4,10). Thoracoscopy may also be used for removal of tracheocele (2,12).

As a conclusion in this asymptomatic case the diagnosis is suspected on chest computed tomographic scan and confirmed by fiberoptic bronchoscopy and conservative treatment is chosen. Although this case is thought to be an acquired one, contrary to the literature, the orifices were small and multiple which is mostly seen in congenital cases.

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