Case Report / Olgu Sunumu

A rare cause of odynophagia: infected tracheal diverticulum

Nadir bir yutkunma güçlüğü nedeni: Enfekte trakeal divertikül

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ABSTRACT

Tracheal diverticulum is a relatively rare lesion. Tracheal diverticula are divided into two subgroups as congenital and acquired with different characteristics and etiologies. The majority of these anomalies is asymptomatic and found as incidental findings on radiological imaging. This article presents a case of an infected tracheal diverticulum presenting with odynophagia. It should be noted that infection may obstruct the air-filled lumen of the diverticulum, causing a diagnostic challenge. Multislice computed tomography with coronal reconstructed images is the modality of choice for diagnosing diverticulum and assessing therapeutic response in associated complications.

Keywords: Computed tomography; diverticulum; trachea.

ÖZ

Trakeal divertikül oldukça nadir bir lezyondur. Trakeal divertiküller farklı özellikleri ve etyolojileri ile doğuştan ve edinsel olmak üzere iki alt gruba ayrılır. Bu anomalilerin çoğunluğu asemptomatiktir ve radyolojik görüntülemede rastlantısal olarak saptanır. Bu yazıda yutkunma güçlüğünün eşlik ettiği bir enfekte trakeal divertikül olgusu sunuldu. Enfeksiyonun hava içeren divertikül lümenini tıkayarak tanısal güçlük yaratabileceği göz önünde bulundurulmalıdır. Koronal rekonstrüksiyon görüntüleri ile çokkesitli bilgisayarlı tomografi divertikül tanısı koymada ve eşlik eden komplikasyonların tedavisine yanıtı değerlendirmede tercih edilmesi gereken yöntemdir.

Anahtar Sözcükler: Bilgisayarlı tomografi; divertikül; trakea.

Tracheal diverticulum is a relatively rare lesion reported in the literature. Rokitansky[1] first described air-containing cysts in the paratracheal region as retention cysts of the mucous glands in 1838. The majority of these abnormalities are asymptomatic and found incidentally on radiological imaging.[2,3] Few cases have been reported in the literature since that original description. We report a case of an infected tracheal diverticulum and discuss the imaging findings for differential diagnosis with paratracheal air-filled lesions.

CASE REPORT

A 27-year-old female, nonsmoker, presented with odynophagia/dysphagia, mild shortness of breath, mild pain when coughing and breathing, without fever for four days. Her medical history was otherwise unremarkable. Physical examination demonstrated soft tissue fullness as well as pain in the right neck area. No mass lesion was palpated. Neck ultrasonography (Figure 1a) showed an ill-defined, heterogeneous hypoechoic mass with

Available online at
www.kbbihtisas.org
doi: 10.5606/kbbihtisas.2015.81205
QR (Quick Response) Code

Received / *Geliş tarihi:* March 02, 2014 Accepted / *Kabul tarihi:* December 12, 2014

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multiple hyperechoic foci at the inferoposterior aspect of the right lobe of the thyroid. Neck and chest computed tomography (CT) demonstrated a hypodense lesion measuring 30x15 mm at the right paratracheal/paraesophageal region, posterolateral to the right tracheal wall just superior to the thoracic inlet (Figure 1b). The trachea and esophagus were displaced to the left by the mass. A narrow connection between the lesion and right posterolateral trachea was noted (Figure 1c, 2a, b). The clinical symptoms of the patient responded to antibiotic treatment. On a follow-up CT, most of the lumen of the lesion was filled with air and the connection with the trachea was more prominent (Figure 1d, e, 2c, d). There was no communication with the digestive tract on a barium-swallow contrast study. The patient refused further diagnostic workup with bronchoscopy and esophagoscopy. The clinical and radiographic follow-up provided the diagnosis of an infected tracheal diverticulum. Surgery was recommended. However, she refused the surgical resection after successful conservative treatment, and appropriate clinical follow-up was suggested.

DISCUSSION

Tracheal diverticula are relatively rare entities. The frequency of the tracheal diverticulum in some autopsy series has been estimated at 1%.[4] On the other hand, their incidence has been reported as 0.75-5.2% with CT.[4-6]

Tracheal diverticula are divided into two subgroups with different characteristics and etiologies: congenital and acquired.[2] A congenital tracheal diverticulum is more common in males than females and considered as a malformed, vestigial, supernumerary branch of the trachea or aborted abnormally high division of the primary lung bud. This malformation may be associated with other upper aerodigestive tract anomalies, such as Mounier-Kuhn disease, tracheoesophageal fistula, and tracheobronchomegaly. The wall of the congenital form resembles that of the trachea, containing smooth muscle fibers and cartilage in addition to the respiratory epithelium. The congenital tracheal diverticulum is smaller than the acquired form and located approximately 4-5 cm below the true vocal cords or just above the carina, usually on the right side of the trachea. The acquired diverticulum is reportedly associated with chronic bronchopulmonary disease. This form is thought to be related to the chronic cough that causes persistent increase in tracheal intraluminal pressure and results from mucosal herniation through weakened

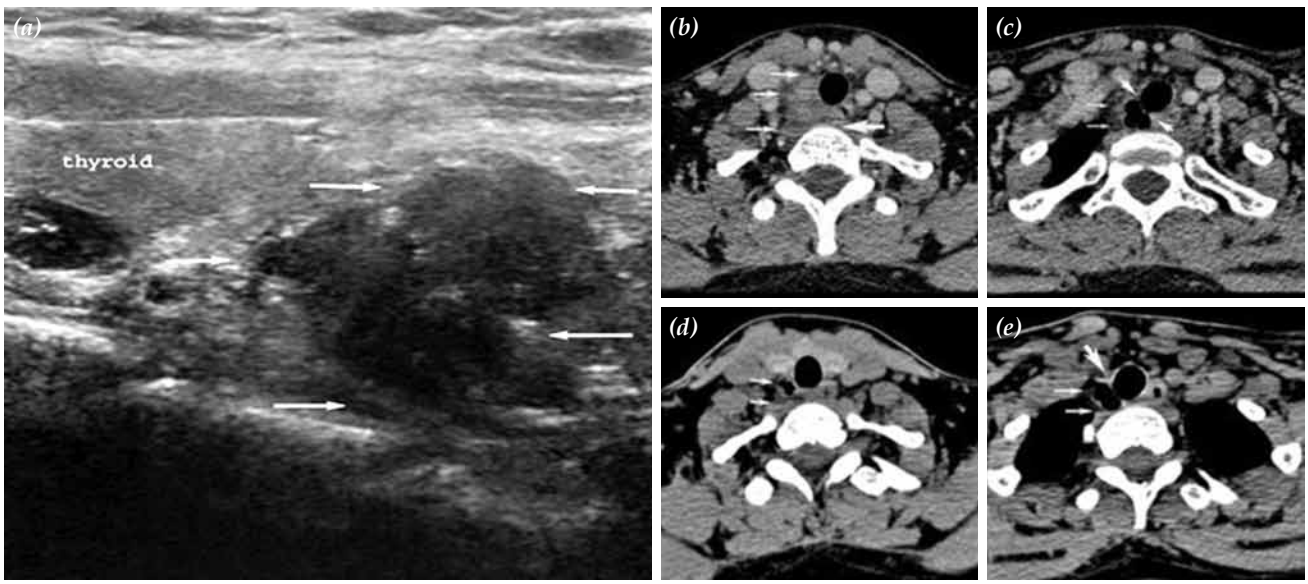

Figure 1. ***(a)*** *Neck ultrasound demonstrates a heterogeneous hypoechoic mass (arrows) with hyperechoic foci at the inferoposterior aspect of the right thyroid lobe.* ***(b)*** *Neck computed tomography shows a hypodense right paratracheal lesion (arrows) displacing trachea and esophagus (big arrow) to the left.* ***(c)*** *Note the air density within the lesion (arrows) and narrow connection between the lesion and trachea (big arrow).* ***(d, e)*** *On a follow-up computed tomography, most of the lumen of the lesion was filled with air and the infected component had regressed. The tracheal cartilage seems to be in continuity by wall of diverticulum (big arrow).*

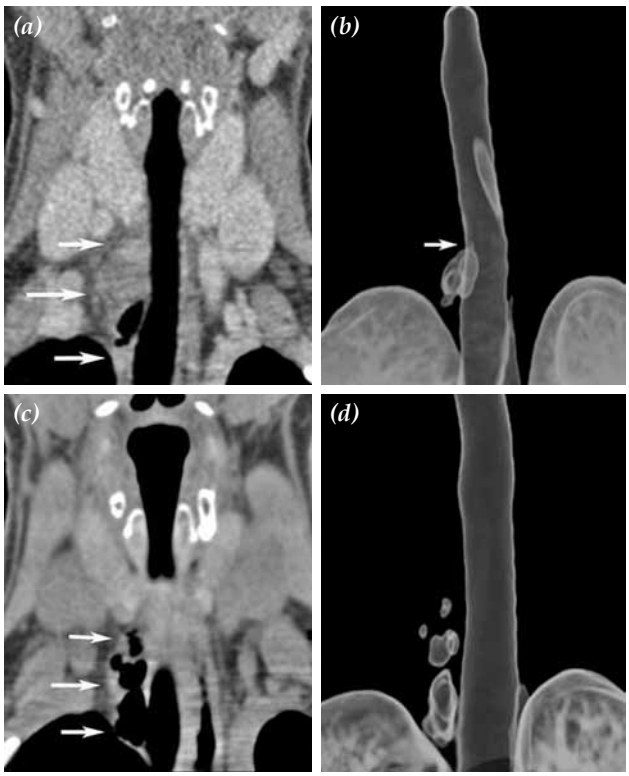

Figure 2. ***(a)*** *Computed tomography with coronal reconstructed image shows the infected paratracheal diverticulum (arrows).* ***(b)*** *Volume-rendered three-dimensional image clearly reveals the narrow connection (arrow) between the trachea and the diverticulum.* ***(c, d)*** *After antibiotic treatment, most of the lumen of the diverticulum was filled with air.*

musculature of the trachea wall due to repeated respiratory infections. Typically, the acquired form has wide connections to the trachea and is larger than the congenital one. This form can appear at any level and consists of only respiratory epithelium and lacks cartilage or smooth muscle.[2,3,7]

Goo et al.[4] found that 98% of tracheal diverticula are located at the right posterior-lateral aspect of the trachea, usually at the level of the thoracic inlet between the T_1 and T_3 vertebra. This fact can be explained by the relative positions of the trachea and esophagus; the supportive effect of the esophagus to the trachea is along its left posterolateral side, leaving the right side of the trachea relatively unsupported.[2,3]

A tracheal diverticulum is commonly an incidental finding on routine radiological investigation and is usually asymptomatic. The symptoms vary with the size and the location of the diverticulum and may result from a local mass effect and direct compression of the trachea, leading to cough, dyspnea, stridor, dysphagia, and chest, neck and/or right clavicle pain. Symptoms can also be related to vagal irritation, resulting in cough, dysphonia, or vocal fold paralysis. Furthermore, it can act as a reservoir for retained secretions and may present with chronic cough, repeated episodes of respiratory infections, hemoptysis, stridor, or dyspnea. In addition, tracheal diverticula have also been detected incidentally during tracheostomy, laryngectomy, unsuccessful intubation or inefficient ventilation, and following trauma.[2,3]

Our case presented with odynophagia/dysphagia due to an infected tracheal diverticulum. A few different presentations of infected tracheal diverticula have been reported in the literature. Teh et al.[3] presented a case of an infected tracheocele which was removed surgically due to persistent and recurrent symptoms. Charest et al.[7] reported an infected acquired tracheocele mimicking malignancy in a patient with a history of lymphoproliferative disease. Also, an infected tracheal diverticulum mimicking a cervical abscess and requiring surgical management has been described.[8]

Goo et al.[4] reported that paratracheal air cysts were visible in only 14% of patients on chest radiographs. At ultrasonography, the paratracheal air cysts appeared as hypoechoic mass-like lesions with multiple echogenic foci, probably due to the strong reflection of the ultrasound beam and reverberation artifacts.[9] Cervical-thoracic CT examination, including thin sections and three-dimensional reconstructed images in the coronal plane, has been shown to be the most effective imaging modality for evaluating the presence and features of tracheal diverticula. On CT scans, the paratracheal diverticulum is characterized by an air-filled tubular structure, often located posteriorly and to the right of the trachea and communicating with the trachea.[2,3,7] Inflammatory changes around the diverticulum may also be visualized. In addition, CT can demonstrate the possible damage of lung parenchyma due to bronchopulmonary disease. It was reported that a CT scan can be helpful to determine whether the lesion

is congenital or acquired by identifying the presence or absence, respectively, of cartilaginous rings in the diverticulum.[2] Chest CT revealed no abnormality in the lung parenchyma in our case. Our patient had no medical history of chronic cough, chronic obstructive pulmonary disease or repeated respiratory infections that could lead to the development of acquired tracheal diverticulum. In our case, the tracheal cartilage seemed to be in continuity with the wall of the tracheal diverticulum (Figure 1e). The connection between the diverticulum and the trachea was narrow which probably resulted in poor drainage of the secretions and subsequent infection. As there were no predisposing factors for an acquired diverticulum, we think that this paratracheal air cyst may be a congenital tracheal diverticulum. But in this case, the diverticulum was larger than the congenital diverticula described in the literature, which may be explained by the infection. However, it is difficult to determine with certainty whether this was a congenital or acquired lesion, since it is not very easy to identify tracheal cartilage in young patients and no pathological evidence is available.

The differential diagnosis of air-filled lesions in the paratracheal region includes lymphoepithelial cyst, bronchogenic cyst, laryngocele, pharyngocele, Zenker's diverticulum, third and fourth branchial anomalies, tracheal diverticula, an apical hernia of the lungs and apical paraseptal bullae or blebs, and pneumomediastinum.[3,4] Radiological barium-swallow contrast study, endoscopic techniques (bronchoscopy and esophagoscopy), and CT can be used to characterize the lesion and exclude tracheal diverticula from the differential diagnoses listed above. Although bronchoscopy can provide definitive diagnosis, diverticula with a narrow opening or just a fibrous connection with the trachea can be bronchoscopically missed. Therefore, multislice CT imaging is the modality of choice for diagnosing diverticula and assessing therapeutic response in associated complications, as shown in our case.

Treatment methods vary throughout the literature, including surgical resection, fulguration, endoscopic cauterization with laser or electrocoagulation, endoscopic division with biopsy forceps, and conservative medical management of symptoms.[2,3] It was suggested that surgery should be considered only in young, symptomatic patients for whom conservative treatment (e.g., antibiotics, mucolytics and physiotherapy) has been unsuccessful.[3] Conservative management was effective in our case. Although surgical treatment was proposed as decided in other cases with infected tracheal diverticula,[3,8] our patient refused surgical resection because of successful conservative treatment. The potential pitfalls secondary to the tracheal diverticulum were discussed with the patient and clinical follow-up was recommended.

Conclusion

This article reports an infrequent symptomatic complication of a rare tracheal malformation. The combination of the radiological findings and effective treatment with antibiotic therapy provided the diagnosis of an infected tracheal diverticulum presenting with odynophagia. The tracheal diverticulum must be considered as a reservoir for secretions that may cause infection. This case emphasizes that an infected tracheal diverticulum should be included in the differential diagnosis of a complicated paratracheal mass. It should be noted that infection remains a diagnostic dilemma by obscuring the air within the lumen of the diverticulum and multislice CT with coronal reconstructed images can be helpful to provide the diagnosis.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

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