

Infected Paratracheal Air Cyst: A Case Report

감염된 기관주위 공기낭: 증례 보고

Hyun Jeong Kim, MD, Sung Shick Jou, MD*, Young Tong Kim, MD, Jong Kyu Han, MD

Department of Radiology, Soonchunhyang University College of Medicine, Cheonan Hospital, Cheonan, Korea

An air-filled paratracheal cyst is a common radiological finding. It may be a congenital defect or an acquired lesion. "Acquired paratracheal cyst" is the term given to the acquired abnormalities, which usually arise in adults. They result from a weakness of the tracheal wall, and they may be caused by trauma, infection, high pressure injuries, long lasting tracheostomy, and obstructive tracheal disease. Majority of the paratracheal air cysts are asymptomatic and are discovered incidentally on radiological images. Also, the management is primarily conservative treatment. Here, we report a case of an infected paratracheal air cyst on the right posterolateral wall of the trachea, which developed into an abscess and was visualized on follow-up multidetector computed tomography and was surgically removed due to persistent symptoms.

Index terms

Paratracheal Air Cyst
Computed Tomography

INTRODUCTION

A paratracheal air cyst is an out-pouching of the trachea lined by respiratory epithelium. The most common location of paratracheal air cysts is the right posterolateral tracheal wall. A paratracheal air cyst results from a weakness of the tracheal wall. The best diagnostic procedures are endoscopic examination and computed tomography (1). Trauma, infection, high pressure injuries, long lasting tracheostomy, and obstructive tracheal disease are the proposed etiologies and these cysts can cause a variety of chronic and recurrent aerodigestive tract symptoms (2).

We report a case of an infected paratracheal air cyst, which revealed progressive abscess formation on follow-up multidetector computed tomography (MDCT) and was ultimately treated by surgical resection.

CASE REPORT

A 53-year-old woman was admitted with complaints of fever,

sore throat, cough, sputum and right neck pain for the past four days. She had no history of any surgical operation and systemic disease. The findings on auscultation of the lungs and physical examination of other organs were normal except for tenderness over the right anterior neck. Routine laboratory test results were unremarkable except for white blood cell count and inflammatory markers. A chest radiograph revealed a round, radiolucent right paratracheal air cyst with a longest diameter of approximately 1.6 cm and tracheal indentation and a thickened right paratracheal stripe (Fig. 1). Both lung fields were unremarkable. Pre-contrast chest MDCT performed on the same day demonstrated an air-filled cyst with fat infiltration in the right posterolateral wall of the trachea at the thoracic inlet level. The cystic lesion measured approximately 2.5 × 2.2 × 1.3 cm in size. On contrast-enhanced chest MDCT performed three years ago, a right paratracheal air cyst was noted without fat infiltration (Fig. 2). Follow-up contrast-enhanced chest MDCT after three days demonstrated that the wall of the cystic lesion was thickened and enhanced (Fig. 3). An air-fluid level was seen within

Received December 12, 2015

Revised January 16, 2016

Accepted March 22, 2016

*Corresponding author: Sung Shick Jou, MD

Department of Radiology, Soonchunhyang University

College of Medicine, Cheonan Hospital,

31 Suncheonhyang 6-gil, Dongnam-gu,

Cheonan 31151, Korea.

Tel. 82-41-570-3515 Fax. 82-41-579-9026

E-mail: ssmri@schmc.ac.kr

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

the cystic lesion. The patient complained of persistent right neck pain after treatment with antibiotics.

In light of her persistent symptoms, a decision was made to perform surgical resection. A right collar neck incision was performed under general anesthesia. A cystic mass with adhesion and inflammatory change was identified and excised. However, a communication with the trachea was not identified due to adhesion.

Grossly, the resected specimen measured $2.0 \times 1.7 \times 0.7$ cm. Histologically, it was confirmed to be a cyst lined by ciliated columnar epithelium, which was consistent with a paratracheal air cyst (Fig. 4).

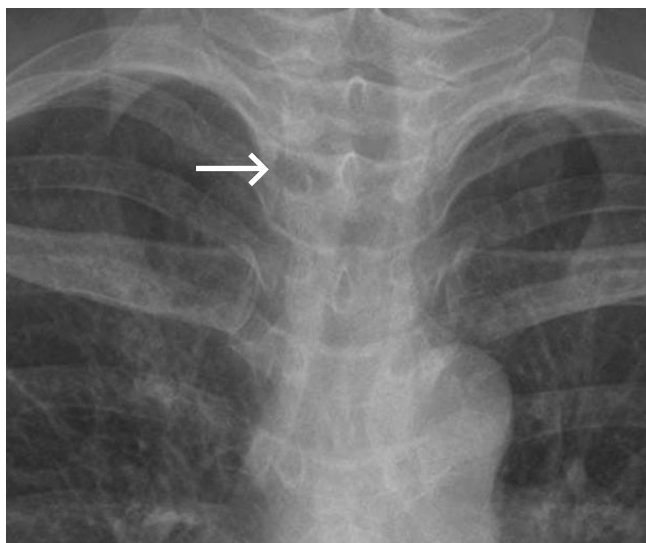


Fig. 1. Infected paratracheal air-cyst. A 53-year-old woman presented with a fever, sore throat, cough, sputum, and right neck pain for the past four days. Chest radiography demonstrates a round, radiolucent right paratracheal lesion (arrow) and right paratracheal stripe thickening.

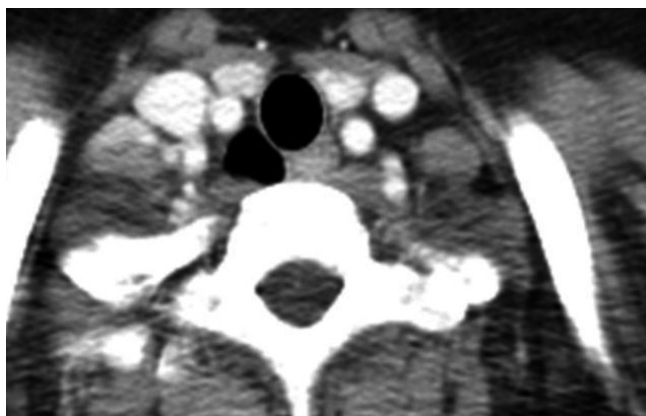


Fig. 2. Contrast-enhanced chest MDCT image obtained three years ago demonstrates an air-filled cystic lesion on the right side of the trachea without fat infiltration. MDCT = multidetector computed tomography

DISCUSSION

Air-filled cysts originating from the trachea are common, whether they are paratracheal air cysts or tracheal diverticula. However, their prevalence is probably underestimated because most cases cause few symptoms and are well tolerated. In a recent radiological study, the prevalence ranged from 0.75% to 6.5% in patients undergoing CT imaging (3, 4).

Conveniently, they are defined as tracheal diverticula and are sub-divided into congenital or acquired lesions. Acquired tracheal diverticula are true diverticula resulting from mucosal herniation through weak points, most commonly in the right posterolateral trachea and usually at the level of the thoracic inlet. They are lined by respiratory epithelium but do not contain cartilage or smooth muscle. Also, they communicate with the trachea (5). Trauma, high pressure injury, and long lasting tracheostomy may be the etiologic factors. In addition to these factors, recurrent infections of the mucous glands of the trachea and chronic cough may play an accessory role in the formation of paratracheal air cysts (2, 6). When these lesions occur at the thoracic inlet level, they are commonly referred to as paratracheal air cysts (6). They have been reported to be associated with pulmonary emphysema, chronic obstructive pulmonary disease, inflammatory conditions such as recurrent pneumonia, chronic bronchitis and bronchiectasis, although their relationship is controversial in the literature (4, 7). Congenital diverticula represent a malformed, vestigial, supernumerary budding of the trachea. The walls of congenital diverticula are similar to the walls of the trachea, containing smooth muscle fibers, cartilage, and respiratory epithelium (8). In addition, they are smaller with narrow, pinpoint openings or pedicles and are located approximately 4–5 cm below the vocal cords or just above the carina. Other malformations, such as a tracheoesophageal fistula, can coexist (5, 8).

Although they are usually discovered incidentally, there are rare reports of paratracheal air cysts causing presenting symptoms. The clinical presentation of the reported cases, usually due to a local mass effect or direct compression of an adjacent structure, includes chronic cough, non-purulent sputum, repeated episodes of respiratory infections, dyspnea, stridor, dysphagia, and chest, neck and/or right clavicular pain (5, 6). Symptoms can also arise due to vagal irritation, retained secretions or recurrent irritation of the upper airways (8). Paratracheal air cysts

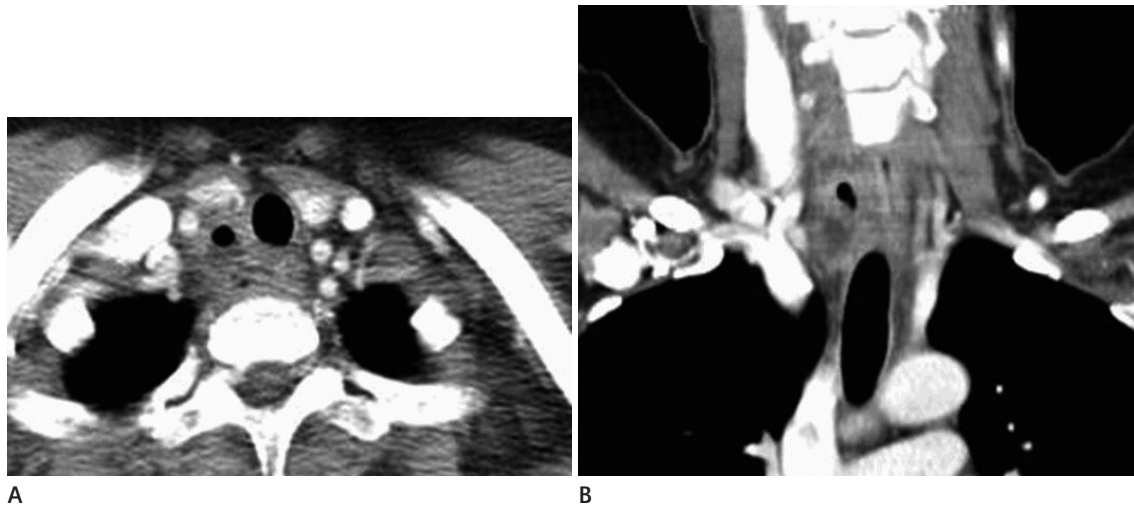


Fig. 3. Contrast-enhanced chest MDCT at present. Axial **(A)** and coronal reformatted **(B)** images demonstrate an air-filled and fluid-filled cystic lesion on the right side of the trachea, measuring about 2.5 × 2.2 × 1.3 cm in size, with an enhancing thick wall due to infection. MDCT = multidetector computed tomography

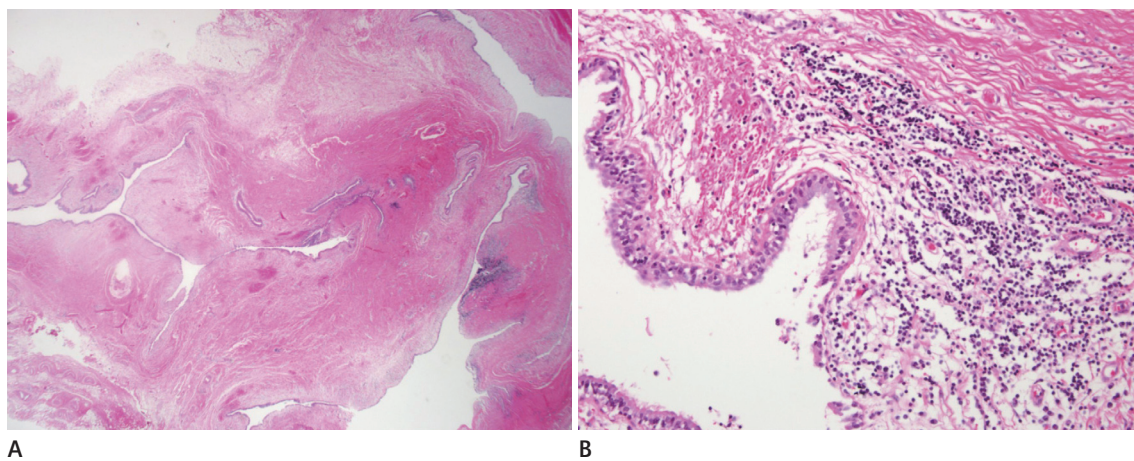


Fig. 4. **A.** Microscopically, the excised mass shows a multilocular cystic appearance in the low power view (hematoxylin and eosin, × 12.5). **B.** The lining cells are ciliated columnar epithelial cells. There is lymphoplasmacytic aggregation in the cystic walls (hematoxylin and eosin, × 200).

have also been found incidentally during tracheostomy, laryngectomy, difficult intubation, and inefficient ventilation (9).

Diagnostic studies include chest radiographs, CT, and bronchoscopy (2).

Goo et al. (10) reported that paratracheal air cysts were visible in only 14% of patients on chest radiographs. On sonography, the paratracheal air cysts appeared as hypoechoic mass-like lesions with reverberation artifacts (5). CT is the most effective method for evaluating the location, origin, size, and features of paratracheal air cysts. It can demonstrate a connection between the cyst and the tracheal lumen. Also, the lung parenchyma can be evaluated (6). Most paratracheal air cysts are located on the right posterolateral aspect of the trachea and at the T1–2 level

on CT. On bronchoscopy, the orifice in the tracheal wall appears as a tiny, well-circumscribed hole. A barium swallow study may be performed to show the relationship with the esophagus, which is usually normal (2).

In our case, the chest radiograph revealed a round, radiolucent right paratracheal air cyst with paratracheal stripe thickening and contrast-enhanced chest MDCT demonstrated an air-filled and fluid-filled cyst with an enhancing thick wall of the right side of the trachea.

The differential diagnosis of an air-filled paratracheal region includes a laryngocele, a pharyngocele, a Zenker’s diverticulum, tracheal diverticula, third and fourth branchial anomalies, para-septal bullae, and apical lung herniations (5, 7). Radiological

barium swallow study, endoscopic techniques, and CT can be used to characterize the lesion. For example, differentiation of Zenker's diverticulum from paratracheal air cyst can often be accomplished by demonstrating a communication between the cyst and the digestive tract or lung apex using barium swallow and axial imaging studies (5).

In symptomatic patients, the approach to treatment is tailored to the patient considering the age and comorbidities.

Most infected paratracheal air cysts can be managed conservatively, but there are few reports of lesions requiring surgical intervention. Surgery is typically reserved for symptomatic lesions that pose a potential health threat (7). It may also be indicated for patients with refractory symptoms. Complete dissection of the paratracheal air cyst and closure of the small communication by a running suture have been performed in symptomatic cases. Thoracoscopy may also be used for the removal of a paratracheal air cyst (2).

In conclusion, infected paratracheal air cysts frequently occur in adults. Although a paratracheal air cyst is commonly asymptomatic and detected incidentally, there are rare reports of paratracheal air cysts presenting with chronic cough, repeated episodes of respiratory infections, hemoptysis, stridor or dyspnea. In our case, the paratracheal air cyst presented as a cervical abscess and caused refractory symptoms despite conservative treatments, such as antibiotics and nonsteroidal anti-inflammatory drugs. In addition, the current case demonstrates that asymptomatic lesions can become problematic over time. Surgery should be considered in these cases. Therefore, it is important to identify lesions using imaging modalities so that the associated abnormalities can be investigated and managed appropriately.

REFERENCES

1. Cheng HM, Chang PY, Chiang KH, Huang HW, Lee CC. Prevalence and characteristics of paratracheal air cysts and their association with emphysema in a general population. *Eur J Radiol* 2012;81:2673-2677
2. Yazkan R, Ozpolat B, Firat H. Tracheocele; a case of rare clinical entity. *Tuberk Toraks* 2008;56:315-318
3. Kosehan D, Kayihan A, Kakteker A. Incidental right paratracheal air cyst: significance of 64-detector multislice CT in differential diagnosis. *New J Med* 2011;28:62-63
4. Boyaci N, Sen Dokumaci D, Karakas E, Yalcin F, Oney Kur-naz AG. Paratracheal air cysts: prevalence and relevance to pulmonary emphysema and bronchiectasis using tho-racic multidetector CT. *Diagn Interv Radiol* 2015;21:42-46
5. Şahin N, Solak A, Genç B. A rare cause of odynophagia: in-fected tracheal diverticulum. *Kulak Burun Bogaz Ihtis Derg* 2015;25:126-130
6. Amaral CB, Silva S, Feijó S. Infected tracheal diverticulum: a rare association with alpha-1 antitrypsin deficiency. *J Bras Pneumol* 2014;40:669-672
7. Choi AR, Choi SH, Kim SW, Sung DW, Rha YH. A case of recurrent respiratory infection resulting from a congenital anomaly of the bronchial tree tracheal bronchus. *Korean J Pediatr* 2008;51:660-664
8. Teh BM, Hall C, Kleid S. Infected tracheocele (acquired tracheal diverticulum): case report and literature review. *J Laryngol Otol* 2011;125:540-545
9. Polat AV, Elmali M, Aydin R, Ozbay A, Celenk C, Murat N. Paratracheal air cysts: prevalence and correlation with lung diseases using multi-detector CT. *J Med Imaging Radiat Oncol* 2014;58:144-148
10. Goo JM, Im JG, Ahn JM, Moon WK, Chung JW, Park JH, et al. Right paratracheal air cysts in the thoracic inlet: clinical and radiologic significance. *AJR Am J Roentgenol* 1999;173:65-70

1. Cheng HM, Chang PY, Chiang KH, Huang HW, Lee CC.

감염된 기관주위 공기낭: 증례 보고

김현정 · 조성식* · 김영통 · 한종규

기관기관지낭은 흔한 영상의학적 소견이다. 이는 선천적 결함 또는 후천적으로도 나타날 수 있다. '후천적 기관기관지낭'은 후천적으로 생긴 경우로 국한되며, 대개 어른에서 발생한다. 이들은 외상, 감염, 고압 손상, 기관절개술의 오랜 유지, 폐쇄성 기관지 질환에 의해 기관지 벽이 약화되어 생긴다. 기관기관지낭의 대부분은 무증상이며, 영상검사에서 우연히 발견된다. 또한, 주로 보존적 치료를 하게 된다. 본 논문에서, 추적관찰을 위해 시행한 다절편 CT에서 농양으로 발전하여 지속적인 증상을 유발하여 수술적으로 제거된 기관지 우측 후외벽의 감염된 기관기관지낭 사례를 보고하고자 한다.

순천향대학교 의과대학 천안병원 영상의학과