Korean J Thorac Cardiovasc Surg 2016;49:313-316 □ CASE REPORT □
ISSN: 2233-601X (Print) ISSN: 2093-6516 (Online) http://dx.doi.org/10.5090/kjtcs.2016.49.4.313

Voice Change Due to Paratracheal Air Cysts

Youn Ju Rhee, M.D.[1], Sung Joon Han, M.D.[1], Yoo Young Chong, M.D.[1], Hyun Jin Cho, M.D.[1], Shin Kwang Kang, M.D., Ph.D.[1], Choong-Sik Lee, M.D.[2], Min-Woong Kang, M.D., Ph.D.[1]

Departments of [1]Thoracic and Cardiovascular Surgery and [2]Pathology, Chungnam National University Hospital, Chungnam National University School of Medicine

Paratracheal air cysts are a rare entity in which cystic formation occurs adjacent to the trachea. Most patients with paratracheal air cysts are asymptomatic, and the cysts are detected incidentally on chest radiograph or computed tomography (CT) scan. Most symptomatic patients complain of pulmonary symptoms or repeated respiratory infection. Rarely, the air cysts can lead to paralysis of the recurrent laryngeal nerve as a result of direct compression. We report a case of a 59-year-old male patient who presented with voice change, and the cause was identified as paratracheal air cysts on a chest CT scan. Surgical resection via video-assisted mediastinoscopy was performed, and the voice recovered immediately after the operation.

Key words: 1. Diverticulum
2. Recurrent laryngeal nerve
3. Mediastinoscopy

Case Report

A 59-year-old man presented with voice change that began 1 month prior. His smoking history was 20 pack-years, and there was no specific past medical history. The patient's review of symptoms was normal except for hoarseness, and there were no abnormal findings on the physical examination, laboratory data, or chest radiography. No abnormal findings were detected on the vocal cords that could cause hoarseness, such as nodules or palsy, on the laryngoscope. The patient underwent a computed tomography (CT) scan, and 3-dimensional (3D) chest CT revealed multiple air-filled lesions in the right paratracheal region (Fig. 1). There was no pulmonary emphysematous lesion on the chest CT. Esophagography was performed to confirm whether there was communication between the esophagus and paratracheal air cyst. However, leakage of contrast was not detected on the examination. There were no orifices in the tracheal wall that suggested a connection between the trachea and the paratracheal air cyst on bronchoscopy. We operated on the patient for the cyst via mediastinoscope. We inserted a video-assisted mediastinoscope into the anterior mediastinum following the crease of a 5.08-cm suprasternal incision and dissected along the anterior surface of the trachea. There were two large, fluid-filled cystic masses on the right side of the trachea, and one mass was compressing the right recurrent laryngeal nerve. We dissected these masses with the assistance of the video mediastinoscope and retrieved the upper pole of the cystic masses through the incision. There was one narrow communication to the right posterior side of the trachea at the thoracic inlet level (Fig. 2).

One of the two masses consisted of a thick-walled

Received: September 1, 2015, Revised: October 4, 2015, Accepted: October 5, 2015, Published online: August 5, 2016

Corresponding author: Min-Woong Kang, Department of Thoracic and Cardiovascular Surgery, Chungnam National University Hospital, Chungnam National University School of Medicine, 282 Munhwa-ro, Jung-gu, Daejeon 35015, Korea
(Tel) 82-42-280-7375 (Fax) 82-42-280-7373 (E-mail) dreamerkang@hanmail.net

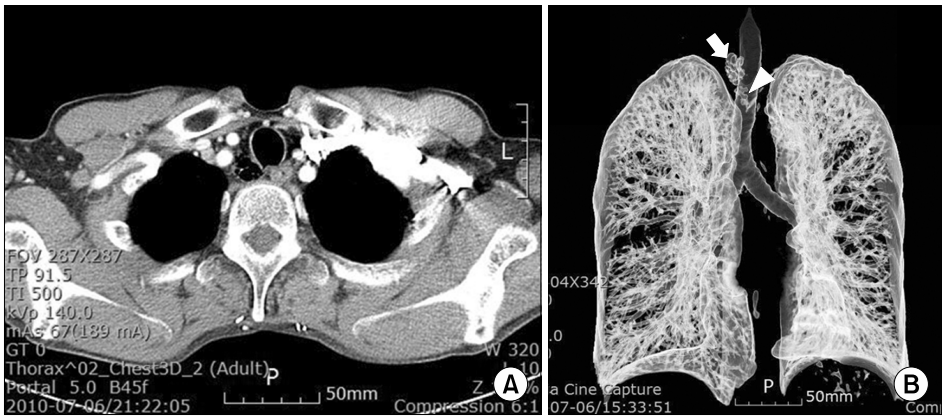

Fig. 1. There are multiple air-filled lesions at the right paratracheal region in (A) axial view and (B) 3-dimensional reconstruction view. Arrow, upper cystic lesion; arrow head, lower cystic lesion.

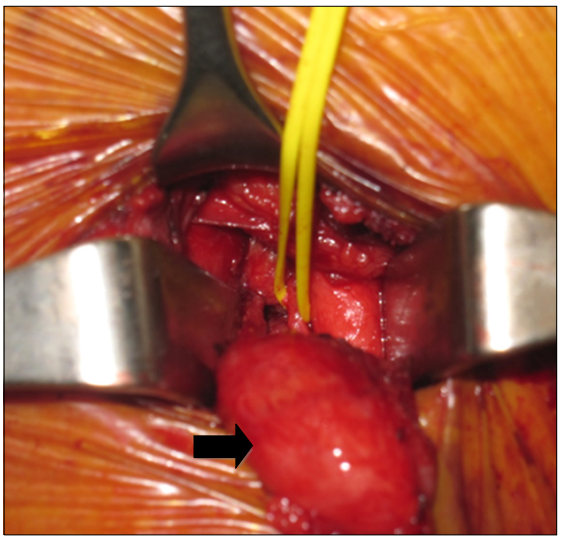

Fig. 2. One of two masses on the right side of the trachea compressed the right recurrent laryngeal nerve snared yellow line (The arrow indicates a cystic lesion).

multilocular cyst, measuring 2.5×2 cm in size. The other cystic mass also was same the mass, measuring 2.5×1.7 cm in size. Histologic findings showed cysts lined by respiratory ciliated columnar epithelium, suggesting tracheal diverticulum (Fig. 3).

Hoarseness had improved by the first day postoperatively. The patient was discharged with no complications on the fifth day postoperatively. The pathology report described the lesions as paratracheal air cysts (Fig. 3). The patient's voice had made a full recovery when he visited the outpatient department for his 1 weeks later after discharge. Three months later, no recurrence was noted.

Discussion

The term paratracheal air cyst is a relatively nonspecific term for a paratracheal air collection. The differential diagnosis of such collections includes tracheal diverticulum, laryngocele, pharyngocele, Zenker's diverticulum, apical hernia of the lung, and apical paraseptal blebs/bullae [1].

Tracheal diverticulum is considered rare, and there is scant published literature for this entity. One study by Goo et al. [1] examined 65 patients with CT evidence of a paratracheal cyst without pathologic differentiation. Nearly all of the cysts were located in the right paratracheal region with only one located in the left paratracheal region. The right-sided nature of the diverticulum may be because the esophagus generally lies to the left of the trachea at this level, leaving the right side unsupported. The majority of the air cysts were at the T2 level. The transverse diameter of the cysts ranged from 5 to 20 mm on the axial scan, and the vertical length ranged from 5 to 25 mm in the coronal or sagittal view. A communicating channel between the air cyst and trachea was found in 5 patients (8%). A total of 31% of the cysts had irregular wall thickening. On respiratory dynamic CT, the cysts distended during forced expiration and contracted during inspiration. A change in the size of the paratracheal air cyst during respiration is indicative of communication between the cyst and the airway. Although it was not pathologically proven, the authors felt that the cysts likely represented tracheal diverticulum [1].

Other causes of paratracheal air collection include laryngoceles, which can be seen on CT as diverticulum of the saccule of the laryngeal ventricle. Paha-

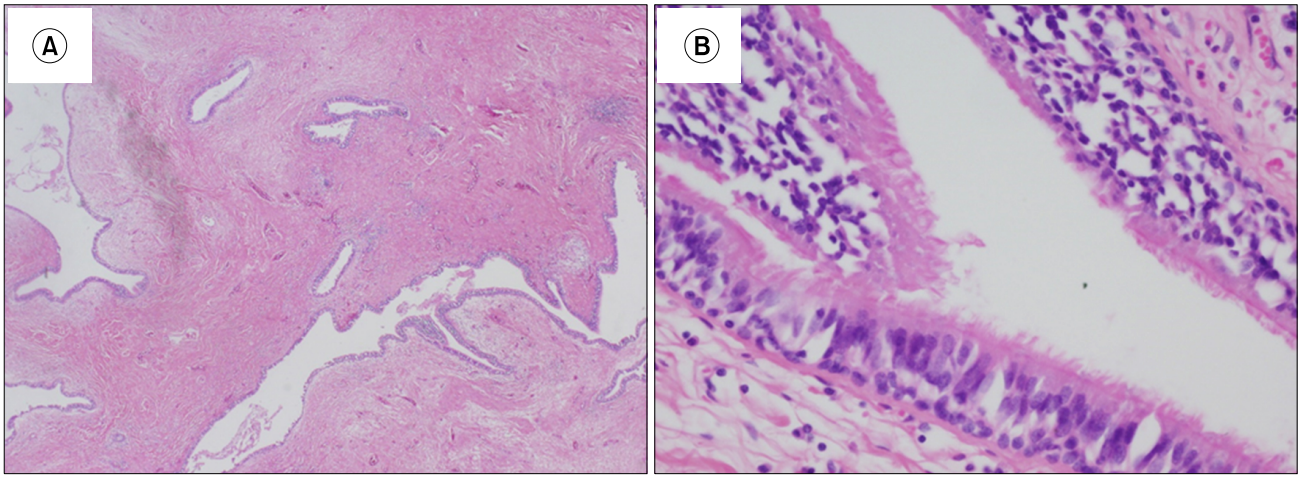

Fig. 3. (A) Many small and large spaces surrounded by dense fibrous tissue are present (H&E, ×12.5). (B) High-power view shows ciliated respiratory epithelium (H&E, ×400).

ryngocele and Zenker's diverticulum can usually be identified by barium examination. An apical hernia has continuity with the body of the lung and has lung markings within the herniated lung on CT scans. Apical paraseptal blebs or bullae, which are air cysts within the lung, can easily be recognized on CT scans [1].

Our patient's paratracheal air cyst had no lung markings, and there was no connection to the lungs or esophagus. A short connection to the trachea was identified on axial images. These findings are typical of tracheal diverticulum.

Specifically, tracheal diverticulum is characterized by single or multiple invaginations of the tracheal wall [1,2]. They are most frequently found incidentally in postmortem examinations, reported in 1% of patients at autopsy [1,2]. Two types of tracheal diverticulum have been described: congenital and acquired. The congenital variety is thought to represent vestigial supernumerary lungs or aborted abnormally high divisions of the primary lung bud. They may arise 4 to 5 cm below the true vocal cords at the right tracheal side or a few cm above the carina. They are relatively small and narrow-mouthed and may occur separately or accompany other congenital anomalies within the tracheobronchial tree, such as tracheoesophageal fistula [3]. The wall of congenital tracheal diverticulum is similar to the actual tracheal wall, containing smooth muscle fibers, cartilage, and respiratory epithelium [4]. The acquired variety is thought to represent an outbulging at a weak spot in the posterior tracheal wall as a result of increased intraluminal pressure, as with a chronic cough [1,2, 4]. In the study by Goo et al. [1], patients with paratracheal air cysts had pulmonary function abnormalities that indicated an obstructive pattern more often than the control group, suggesting a possible association of tracheal diverticuli with emphysema. Acquired tracheal diverticulum can arise at any level, and they are typically wide-mouthed and larger than congenital diverticulum [2,4]. Histologically, they are lined by respiratory epithelium; however, no smooth muscle or cartilage is found in the wall. They may be single or multiple [4].

The diagnosis of tracheal diverticulum is relatively straightforward and can be made radiographically. It is an air-filled tubular structure, most often found posterior and slightly to the right of the trachea and communicating with the trachea. Cartilaginous rings within the wall of the diverticulum suggest a congenital form, whereas the absence of cartilaginous rings may suggest an acquired form [2,5]. CT may also reveal whether the neck of the diverticulum is small or large and whether its wall is thickened as a result of repeated inflammation [5,6]. When the neck of the diverticulum is very small, communication with the trachea may not be clearly visible on CT [2].

Patients with tracheal diverticulum are usually asymptomatic and tend to be incidentally detected; however, tracheal diverticulum can act as a cavity filled with secretions and so may present clinically with chronic cough, dyspnea, stridor, and repeated

episodes of respiratory infection [2,5]. Rarely, tracheal diverticulum can present with symptoms resulting from direct compression of the adjacent tissue or organs [4].

Treatment options in tracheal diverticulum include surgical resection in young or symptomatic patients and conservative medical treatment with antibiotics, mucolytics, bronchodilators, and physiotherapy in the elderly and debilitated [2,4,5]. In our case, because the patient had hoarseness owing to the mass effect of the paratracheal air cyst and was young, surgical treatment was the appropriate decision.

In this case, the patient presented with hoarseness owing to the mass effect of paratracheal diverticulum. During the work-up, there were no significant findings or causes of hoarseness on the laryngoscope. Findings on the chest CT scan and 3D reconstruction demonstrated paratracheal air cyst with a channel between the cysts and the trachea. Air collection located at the right posterior side of the trachea at the thoracic inlet with fistula was a specific finding that suggested tracheal diverticulum. The disease is rare, but it was not difficult to diagnose. Because the patient had a symptom and was young and healthy enough to undergo general anesthesia and surgery, the air cysts were entirely removed via video-assisted mediastinoscopy. Reported methods of approach were thoracotomy or video-assisted thoracoscope, but in a small number of cases, resection of the tracheal diverticulum was performed through video-assisted mediastinoscopy in the literature. In our case, the cysts were excised completely through video-assisted mediastinoscopy. Most tracheal diverticula are located in the thoracic inlet, and therefore, an approach via mediastinoscopy is possible. The air cysts were confirmed as tracheal diverticula on the histologic examination. Immediately after the operation, the hoarseness disappeared.

In conclusion, tracheal diverticulum is a rare disease that presents as an outpouching of the tracheal wall [1]. Most patients with tracheal diverticulum are asymptomatic but can present with respiratory symptoms, repeated respiratory infection or a mass effect [2,5]. Through chest CT, the physician can obtain a great deal of information such as location, size, air cyst wall thickness, and association with adjacent tissue. Depending on the findings, it may be possible to diagnose paratracheal air cyst without histologic examination. If the air cysts do not cause problems, asymptomatic, elderly, or debilitated patients can be treated with conservative management depending on symptoms [2,4,5]. However, if symptoms result from direct compression, surgical resection may be an immediate response to the symptoms.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

References

1. Goo JM, Im JG, Ahn JM, et al. *Right paratracheal air cysts in the thoracic inlet: clinical and radiologic significance*. AJR Am J Roentgenol 1999;173:65-70.
2. Haghi Z, Towhidi M, Fattahi H, Lari SM. *Right paratracheal air cyst (tracheal diverticulum)*. Respir Care 2009;54:1409-11.
3. Shah M, Joshi JM. *Tracheal diverticulum*. Indian J Chest Dis Allied Sci 2012;54:39-40.
4. Sato T, Sasaki Y, Yamasaki M, et al. *A right paratracheal air cyst caused by tracheal diverticula*. Intern Med 2010;49:315-9.
5. Soto-Hurtado EJ, Penuela-Ruiz L, Rivera-Sanchez I, Torres-Jimenez J. *Tracheal diverticulum: a review of the literature*. Lung 2006;184:303-7.
6. Buterbaugh JE, Erly WK. *Paratracheal air cysts: a common finding on routine CT examinations of the cervical spine and neck that may mimic pneumomediastinum in patients with traumatic injuries*. AJNR Am J Neuroradiol 2008;29:1218-21.