

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Percutaneous sclerotherapy for mediastinal cyst resulting in dyspnea due to tracheal compression in an elderly patient: A case report [☆]

Jong Soo Shin, MD, Ph.D.^a, Yeon Soo Kim, MD, Ph.D.^{b,*}

^a Department of Radiology, Hallym University Dongtan Sacred Heart Hospital, 7 Keunjaebong-gil, Dongtan 1(il)-dong, Hwaseong-si, Gyeonggi-do, South Korea

^b Department of Thoracic and cardiovascular surgery, Inje University Ilsan Paik Hospital, 170 Juwaha-ro, Ilsansu-gu, Goyang-si, Gyeonggi-Do, South Korea

ARTICLE INFO

Article history:

Received 10 April 2020

Revised 8 June 2020

Accepted 8 June 2020

Keywords:

Bronchogenic Cyst

Ultrasonography

Fluoroscopy

Sclerotherapy

ABSTRACT

Surgical resection is usually indicated for symptomatic mediastinal tumors. However, surgery potentially increases postoperative complications and hospitalization length in patients who are elderly, in poor general condition, or have tumors located in the thoracic inlet. We present an 84-year-old female with progressive cough and dyspnea for 1 week. Simple radiogram and computed tomography scan showed a large superior mediastinal cyst, sized 8.3 × 6.1 × 4.6 cm, narrowing the trachea. Ultrasonography- and fluoroscopy-guided percutaneous sclerotherapy using alcohol and bleomycin was applied a single time. The patient was discharged 2 days later without complication, and she did not develop symptoms over a 6-year period. Percutaneous sclerotherapy, especially in patients who are elderly or in poor general condition, could be an effective and reliable tool for cyst management.

© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license.

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

Mediastinal cysts are usually asymptomatic masses in adults. Symptoms, if present, result from infection or compression of adjacent structures by cysts [1,2]. Complete surgical resection has been considered the first treatment option for symptomatic cyst [3]. However, a surgical operation is an invasive procedure and requires general anesthesia and relatively long-term hospitalization. In addition, surgery may also result

in various complications depending on the site of the tumor. Needle aspiration for bronchogenic cysts was reported as an alternative treatment, but cysts tend to recur after simple aspiration without sclerotherapy due to active lining cells [3–5].

A variety of benign cystic diseases is currently being treated by sclerotherapy, as it is an effective and less invasive method [6–8]. However, percutaneous needle aspiration and sclerotherapy for treatment of bronchogenic cysts has been rarely reported [3–5]. In the present case, the cyst was palpable, so we could use ultrasonography (US) for

[☆] Conflict of interest: The authors declare that they have no conflict of interest.

* Corresponding author.

E-mail address: kimyns@paik.ac.kr (Y.S. Kim).

<https://doi.org/10.1016/j.radcr.2020.06.021>

1930-0433/© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license. (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

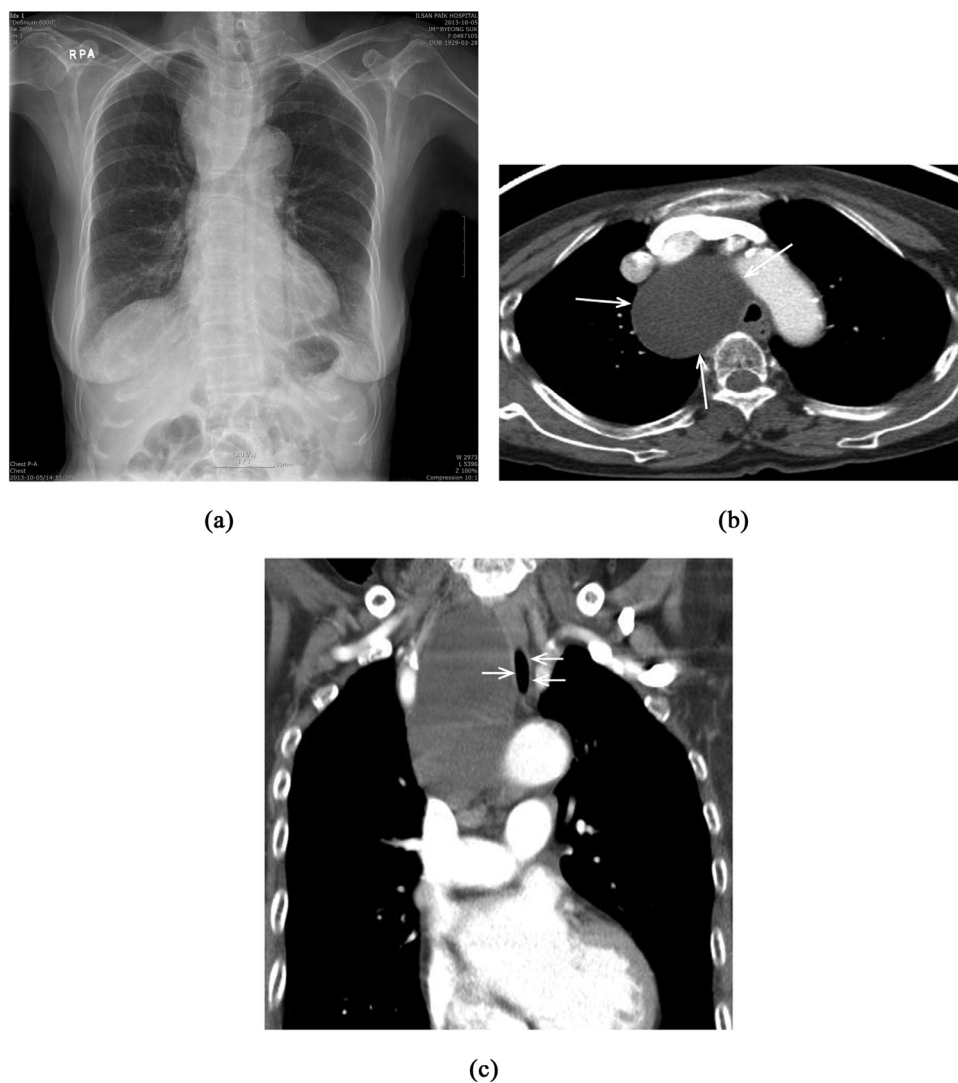


Fig. 1 – Simple radiogram and contrast-enhanced chest computed tomography (CT) scan of an 84-year-old woman with persistent cough and dyspnea. **A:** Chest radiogram shows right mediastinal tumor causing shifting and narrowing of the trachea. **B:** Enhanced axial CT image shows a well-circumscribed, 8.3 × 6.1 × 4.6 cm-sized, nonenhancing, mediastinal cystic mass (arrows) at the right paratracheal area. **C:** Enhanced coronal CT image shows an oval-shaped mediastinal mass at the right paratracheal area with left-side deviation of the trachea (arrows).

localization. Herein, we report percutaneous sclerotherapy with ethanol and bleomycin as the sclerosing agents for successful treatment of a symptomatic mediastinal cyst in an 84-year-old woman.

Case Report

An 84-year-old woman presented to the emergency department for persistent cough and dyspnea that was progressively worsening over the past week. Her past medical history included essential hypertension and rheumatoid arthritis for which she had been treated with medications for decades. Simple radiogram showed a large paratracheal mediastinal tumor that was shifting and narrowing the trachea (Fig. 1A). Enhanced chest computed tomography (CT) images

showed a well-circumscribed, oval-shaped, low-attenuated (25 Hounsfield units), nonenhancing, thin-walled, mediastinal cystic mass without calcification in the right paratracheal area. The size of the cystic mass was 8.3 × 6.1 × 4.6 cm (Fig. 1B), and it was narrowing and shifting the trachea to the left side (Fig. 1C). Although surgical resection was considered, percutaneous sclerotherapy was planned as the initial treatment and diagnostic trial for the mediastinal mass given the patient's old age and location and benign nature of the tumor. The procedure was explained to the patient, who provided written consent.

The patient was placed in the supine position, and local anesthesia was performed with 2% lidocaine hydrochloride at the puncture site in the right supraclavicular region. Under US guidance, the mediastinal cystic mass was punctured with a 21-gauge Chiba needle (Fig. 2A). A 5-French (Fr) Kumpe catheter (Cook, Bloomington, IN, USA) was inserted within the

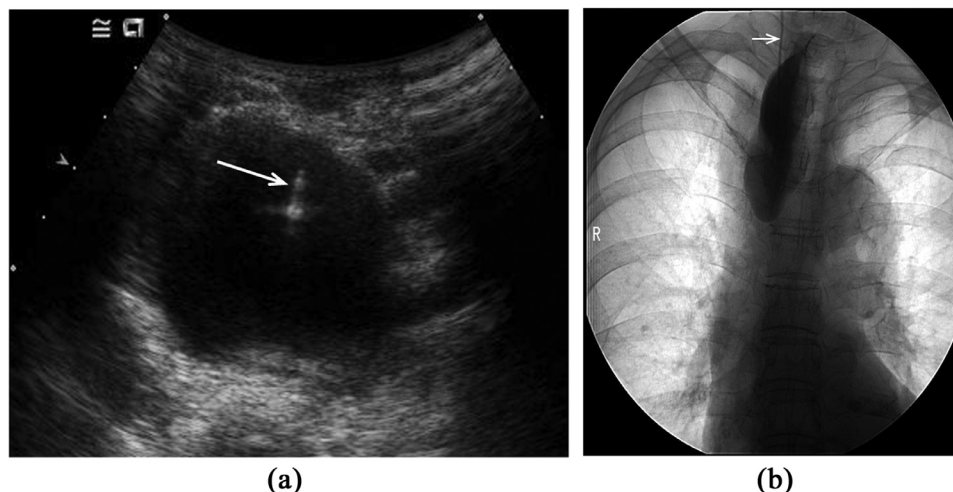


Fig. 2 – Percutaneous ultrasonography (US)- and fluoroscopy-guided sclerotherapy for bronchogenic cyst. A: The cystic lesion was punctured with a 21-gauge needle (arrow) under US guidance. The cystic fluid was completely aspirated. B: A mixture of bleomycin, ethanol, and contrast media was injected through a 5-Fr catheter (arrows) under fluoroscopy guidance. There was no contrast leakage around the mediastinal mass.

cyst using a guide wire under fluoroscopic guidance (Multistar Plus, Siemens, Erlangen, Germany). The cystic fluid was aspirated as completely as possible and was sent to a laboratory for cytology. The total amount of aspirated brown, mucinous fluid was 140 mL. The cyst was then filled with 30 mL of a 10%-diluted, water-soluble contrast media. After confirming that no contrast fluid had leaked from the cyst, we completely aspirated the diluted contrast media. A mixture of 15 mL of bleomycin (15 mg of bleomycin dissolved in 15 mL of normal saline), 10 mL of 99% absolute ethanol, and 5 mL of contrast media were injected through the 5-Fr catheter (Fig. 2B). Then, the patient was placed in the prone, supine, and lateral decubitus positions for 30 minutes to allow adequate contact of the sclerotic agent with the wall of the cyst. The mixture of bleomycin, absolute ethanol, and contrast media was then removed, followed by the catheter. Histopathologic analysis of the aspirate material showed no malignant cells. Because of the mucoid nature of the fluid, the lesion was suspicious for a bronchogenic cyst.

Immediately after the procedure, the patient's respiratory symptoms resolved, and she was discharged 2 days later. After 1 month, follow-up chest CT scan showed nearly complete regression of the cyst with improvement of tracheal deviation (Fig. 3A, B). Six years later, the patient reported no symptoms, and there was no tumor recurrence on a simple radiogram (Fig. 4).

Discussion

A bronchogenic cyst is a rare, benign, congenital, cystic lesion that develops from abnormal budding of the tracheal diverticula during the embryologic period. Bronchogenic cysts are usually located in the mediastinum [2]. The cyst wall contains columnar respiratory epithelium, mucous glands,

cartilage, smooth muscle tissue, and fibrous tissue [2,5]. The fluid within bronchogenic cysts is usually a mixture of water and proteinaceous mucus [1,2,5].

On CT images, bronchogenic cyst appears as a round or elliptical cystic mass with a well-defined, thin, smooth wall that is not contrast enhanced [1,5]. If the lesion demonstrates soft tissue density, differentiating the cyst from lymph nodes or other solid lesions is difficult [1,3]. On magnetic resonance imaging, bronchogenic cysts frequently show higher signal intensity on T1- and T2-weighted images than that of muscle. However, T1-weighted magnetic resonance imaging may show variable patterns of signal intensity because of variable cystic contents and presence of protein, hemorrhage, or mucoid material [1,3].

Sclerotherapy has been used as a first-line treatment for various benign cysts, such as renal cysts, neck cysts of lymphangioma, branchial cleft cysts, thyroglossal duct cysts, and ranula. It shows good results and offers the advantages of no external scarring, little morbidity, and few complications [6-8]. Sclerotherapy for treatment of bronchogenic cysts was previously reported in a few cases. Johnston et al. [4], Lakadamyali et al. [5], and Li et al. [3] reported CT-guided aspiration and sclerotherapy for bronchogenic cysts, with good results. In our case, the cyst was easily seen on an US exam, so we approached the cyst under US guidance in the same way as other benign cyst ablations, such as renal or neck cyst. Because real-time US- and fluoroscopy-guided sclerotherapy is safer and more accurate than CT-guided sclerotherapy, it is the preferred method if US access is possible.

The optimum agent for bronchogenic sclerotherapy remains to be determined. Johnston et al. [4] used bleomycin and alcohol, Lakadamyali et al. [5] used alcohol, and Li et al. [3] used bleomycin as the sclerosing agent. In our case, we used a mixture of bleomycin and ethanol. Ethanol is the most widely used sclerosing agent for sclerotherapy because it is a potent and less expensive sclerosant [6,9]. Its mechanisms of action include rapid dehydration of the cyst wall, protein

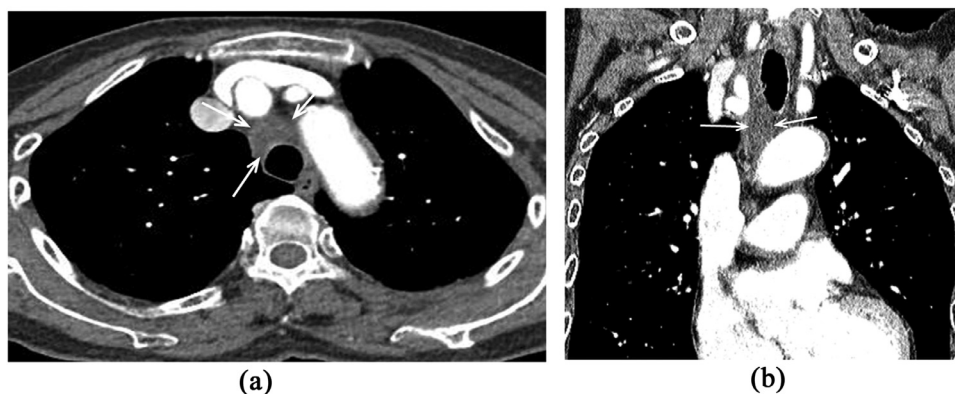


Fig. 3 – One-month follow-up, contrast-enhanced chest CT scan. A. Enhanced axial chest CT image shows that the mediastinal cystic mass was markedly decreased in size (2.0 × 1.4 × 2.5 cm) and volume (arrows). B. Enhanced coronal CT image shows regression of the mediastinal cystic mass (arrows) and marked improvement of left-side deviation of the trachea.



Fig. 4 – Chest radiogram 6 years after sclerotherapy.

denaturation that results in clumping of blood cells, and vessel wall necrosis that results in thrombosis and occlusion [6,8,9]. The potential side effects of ethanol sclerotherapy are nerve injury, skin necrosis, hemoglobinuria, and severe hypotension [8,9]. Therefore, special care must be taken to avoid leakage during injection of ethanol. Bleomycin was first developed as an antitumor agent in 1966, and its mechanism of action was inhibition of DNA synthesis [10]. This drug was also known to produce a sclerosing effect due to its direct action on endothelial cells, producing a nonspecific inflammatory reaction [10]. Bleomycin's sclerosing effect is achieved by local action, which depends on availability of

drug per unit surface area of lesion [3,8]. However, bleomycin as a sclerosing agent must be administered very carefully because it can cause pulmonary fibrosis [3,8].

In conclusion, bronchogenic cysts are benign cystic lesions, but when patients present with symptoms, such as mass effect on surrounding structures, treatment is mandatory. If US access of the lesion is possible, percutaneous US- and fluoroscopic-guided sclerotherapy may be an effective, safe, and minimally invasive treatment method.

REFERENCES

- [1] McAdams HP, Kirejczyk WM, Rosado-de-Christenson ML, Matsumoto S. Bronchogenic cyst: Imaging features with clinical and histopathologic correlation. *Radiology* 2000;217(2):441–6.
- [2] Limaïem F, Ayadi-Kaddour A, Djilani H, Kilani T, El Mezni F. Pulmonary and mediastinal bronchogenic cysts: a clinicopathologic study of 33 cases. *Lung* 2008;186(1):55–61.
- [3] Li L, Zeng XQ, Li YH. CT-guided percutaneous large-needle aspiration and bleomycin sclerotherapy for bronchogenic cyst: report of four cases. *J Vasc Interv Radiol* 2010;21(7):1045–9.
- [4] Johnston SR, Adam A, Allison DJ, Smith P, Ind PW. Recurrent respiratory obstruction from a mediastinal bronchogenic cyst. *Thorax* 1992;47(8):660–2.
- [5] Lakadamyali H, Ergun T, Lakadamyali H, Oguzkurt L. Alcohol ablation therapy of an atypically located symptomatic bronchogenic cyst: a case report. *Cardiovasc Intervent Radiol* 2007;30(6):1274–6.
- [6] Egilmez H, Gok V, Oztoprak I, et al. Comparison of CT-guided sclerotherapy with using 95% ethanol and 20% hypertonic saline for managing simple renal cyst. *Korean J Radiol* 2007;8(6):512–19.
- [7] Kim MG, Lee NH, Ban JH, Lee KC, Jin SM, Lee SH. Sclerotherapy of brachial cleft cysts using OK-432. *Otolaryngol Head Neck Surg* 2009;141(3):329–34.
- [8] Wiegand S, Eivazi B, Zimmermann AP, Sesterhenn AM, Werner JA. Sclerotherapy of lymphangiomas of the head and neck. *Head Neck* 2011;33(11):1649–55.

- [9] Gelczer RK, Charboneau JW, Hussain S, Brown DL. Complications of percutaneous ethanol ablation. *J Ultrasound Med* 1998;17(8):531–3.
- [10] Kumar V, Kumar P, Pandey A, et al. Intralesional bleomycin in lymphangioma: an effective and safe non-operative modality of treatment. *J Cutan Aesthet Surg* 2012;5(2):133–6.