

tion of the ducts due to inflammatory cells, desquamated epithelium, or submucosal fibrosis, or a combination thereof [4, 6]. Other reports demonstrated that the cause of esophageal intramural pseudodiverticulosis may be due to abnormal motor activity of the esophagus, more specifically, the incidence of esophageal intramural pseudodiverticulosis associated with achalasia or the esophageal web has been increasingly cited [6].

Radiographic evaluation is vital in the diagnosis of esophageal intramural pseudodiverticulosis. Barium esophagogram will show numerous tiny flask or collar button-shaped outpouchings consistent with pseudodiverticula, whereas computed tomography in esophageal intramural pseudodiverticulosis is marked by thickening of the esophageal wall, diffuse irregularity of the esophageal lumen, and intramural gas collection [7]. Endoscopy allows direct visualization of the diverticular orifices; however, it has been reported that this is seen in only 20% of patients [5]. The results in our case are well correlated with these findings, and led us to distinctly confirm the diagnosis of esophageal intramural pseudodiverticulosis. Esophageal intramural pseudodiverticulosis has a benign course with a good response to empiric therapy, including anti-inflammatory medication and endoscopic dilation therapy. However, some reports have shown severe complications with esophageal intramural pseudodiverticulosis, including mediastinitis from a fistula or esophageal perforation [2]. In addition, iatrogenic injury from endoscopic manipulation can lead to small localized perforations that affect the anterior mediastinum [8]. Although endoscopic dilation therapy may be necessary in patients with esophageal stricture, the risk of causing lacerations from forced dilation is high and should only be performed by highly experienced endoscopists. On the basis of the previously mentioned findings and due to significant symptoms, our patient underwent dilation carefully using a balloon dilator. Dysphagia improved dramatically without any complications; however, the tiny diverticula did not resolve after treatment.

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Cerebral Air Embolism After Intrathoracic Anti-Cancer Drug Administration

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We report a case of cerebral arterial air embolism that was followed by a brain computed tomographic scan and magnetic resonance imaging during the first week after onset. A 73-year-old man was admitted for treatment of pleural dissemination that was a recurrence after right lower bilectomy for advanced lung cancer. Thirty minutes after an anti-drug administration through the chest drainage tube, he lost consciousness shortly after coughing. A bubble in the inferior sagittal sinus was observed on the day of the stroke, which then disappeared within 24 hours. It seems that the anti-cancer agent evoked inflammation at the visceral pleura and the subject inhaled massive air flow into the systemic circulation.

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Some invasive clinical procedures have a risk of causing air embolism if the visceral pleura is somehow injured. Air embolism was previously recognized as a rare complication of percutaneous transthoracic needle biopsy. Recently, arterial air embolisms such as myocardial and cerebral infarction have also been reported in computed tomographic (CT)-guided marking with a diagnosis of early peripheral lung cancer [1]. We experienced a cerebral air embolism after an anti-cancer drug administration to the pleural cavity and observed the process of cerebral infarction on the findings of a brain CT scan and magnetic resonance imaging.

A 73-year-old man was admitted 2 months after right lower bilectomy and mediastinal lymph node dissection for primary lung squamous cell carcinoma with pulmonary metastasis. A chest tube was inserted into the right pleural cavity for drainage of effusion due to pleural dissemination, which presumably already existed at surgery. To control the pleural effusion, 130 mg of cisplatin and then 10^9 crushed cells of *Streptococcus pyogenes*

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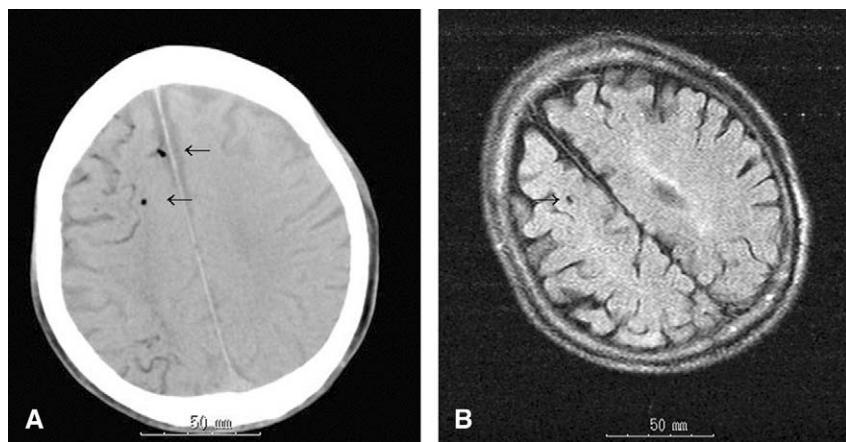


Fig 1. Early phase after the cerebral air embolism. (A) Several bubbles (arrows) can be seen in the inferior sagittal sinus and the superficial cerebral vessels 1 hour after the stroke on a brain computed tomographic scan. (B) Almost all bubbles disappeared 210 minutes after onset on a brain magnetic resonance image. Only one bubble (arrow) can be observed on the anterior cerebral lobe.

treated by penicillin (Picibanil [Chugai Pharmaceutical Co Ltd, Tokyo, Japan]) were slowly injected through the chest tube by manual syringe in a supine position. The tube was kept clamped after the injection. Thirty minutes after administration of the drug the subject lost consciousness shortly after coughing. A brain CT scan revealed a marked bubble sign in the inferior sagittal sinus and small superficial vessels 1 hour after the stroke (Fig 1A). Almost the entire bubble had disappeared and little air was observed on magnetic resonance imaging 210 minutes after the stroke (Fig 1B). The day after the onset, the subject recovered consciousness; however left-sided hemiplegia appeared. This condition continued until the subject died from pneumonia 33 days after onset. No bubble was observed in the great vessels or the heart on a chest CT scan taken 24 hours after the stroke. Subsequently, a high intensity signal appeared at the border

region of the anterior and middle cerebral artery on a T2-enhanced magnetic resonance image 24 hours after the stroke (Fig 2A). This signal was considered to show brain ischemia and edema. Six days after the stroke, the cerebral sulcus and cortex became unclear in the same area on CT scan; the finding was diagnosed as a cerebral infarction (Fig 2B).

Comment

Several decades ago, air embolism was observed to occur during artificial pneumothorax therapy for tuberculosis. Recently cerebral air embolism has rarely been associated with CT-guided needle biopsy and hook-wire localization for minute pulmonary tumors. Morbidity of air embolism caused by percutaneous transthoracic needle biopsy is reported to be 0.07% [2]. Mortality associated

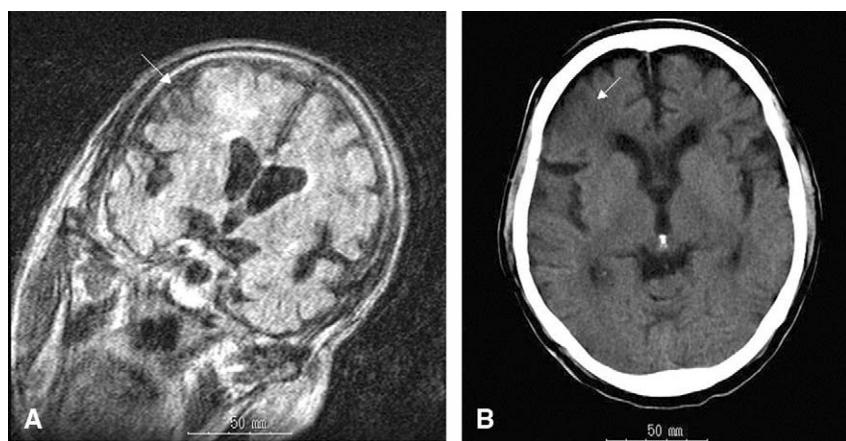


Fig 2. Late phase after the cerebral embolism. (A) After the bubble disappeared, an area of high intensity appeared at the border area between the anterior and middle cerebral arteries (arrows) 24 hours after the stroke on a brain magnetic resonance image. This finding seemed to represent brain ischemia and edema. (B) The cerebral sulcus and cortex became unclear at the same area on a computed tomographic scan (arrow) 6 days after the stroke, representing a cerebral infarction.

with transthoracic needle biopsy or marking of the lung is estimated at 0.002% [3]. This case seems to be the first report of a cerebral air embolism developing after anti-cancer agent administration through a chest tube.

The first clinical characteristic of a cerebral air embolism is that it occurs at the right side of the cerebral area because air can easily flow into the first branch of the aortic arch, which is the right brachiocephalic artery. Afterward, the bubble is usually observed to disappear within 24 hours on brain CT scan or magnetic resonance imaging. In almost all cases in the literature, the subject recovered neurogenic disturbance. However, in our case, a left-sided hemiplegia continued until death, although consciousness returned within 24 hours. Changes in the findings on CT scan and magnetic resonance imaging well reflected the clinical sequence from the detection of the bubble to the development of the ischemia, edema, and infarction. When such a cerebral air embolism occurs, the patient's head should be lowered as quickly as possible under oxygen supply. A hyperbaric oxygen unit should be transferred if possible [4].

Two mechanisms are hypothesized to explain why a large amount of air existed in the peripheral pulmonary vein and then in the systemic circulation. First there was a possibility that the tip of the chest tube that directly contacted the visceral pleura had advanced further into the pulmonary tissue. Of course it was ensured that the tip of the tube was outside the lung when drainage was done. Next the inflammation may be induced by the anti-cancer agent to the visceral pleura [5]. The inflammation may have formed a fistula between the peripheral bronchus and the pulmonary vein. The increased pressure gradient caused a great deal of air to be inhaled into the pulmonary vein through the fistula by means of coughing, and the air then entered the systemic circulation. The second mechanism seems most likely in our case, because air was never expelled from the chest tube during the entire drainage period. Care must be taken to be sure that the tip of the chest should not vertically contact the visceral pulmonary pleura when drugs are scheduled to be administered. A dose of drug should be determined with caution, because the concentration of drugs would markedly increase in a limited space such as a pleural cavity with adhesion.

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Diagnosis of Birt-Hogg-Dube Syndrome in a Patient With Spontaneous Pneumothorax

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Birt-Hogg-Dube syndrome refers to a dermatologic syndrome, consisting of small papular skin lesions distributed on the scalp, forehead, face and neck, which is autosomally dominantly inherited. Subsequently patients may develop concomitant renal and thoracic pathology. We report the case of a patient with Birt-Hogg-Dube syndrome diagnosed after spontaneous pneumothorax.

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The syndrome was reported for the first time in 1977 by Birt, Hogg, and Dube [1] who described small papular skin lesions distributed on the scalp, forehead, face, and neck in 15 of 70 members of the same family. The papula lesions develop after the age of 25 years, are autosomally dominantly inherited, and contain ectodermal and mesodermal components. Histologic examination of the lesions reveals fibrofolliculomas, trichodiscomas, and acrochordons. Subsequently patients may develop concomitant renal carcinoma and bullous lung diseases (eg, lung cysts or bullous emphysema), which are associated with recurrent pneumothorax.

A 43-year-old woman presented with a history of progressive dyspnea for 6 months. Chest roentgenogram revealed a left pneumothorax that was treated with drainage. Thoracic computed tomographic scan showed multiple bilateral cystic lesions of the lung (Fig 1). Family anamnesis revealed a Birt-Hogg-Dube syndrome (BHDS) in a member of her family. The patient indeed had papulose lesions on her face as described in the literature (Fig 2). Histologic examination of one of them revealed a fibrofolliculoma. The patient underwent a left thoracoscopy with wedge resections of the pulmonary cystic lesions for persistent pneumothorax (Fig 3). Histologic assessment revealed benign cysts and the absence of lymphangioleiomyomatosis, which was one of the differential diagnosis revealed by computed tomographic scan images.

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