

Images

# In-flight arterial gas emboli from a ruptured bronchogenic cyst

Edward Mak, BSN\*; Ka Wai Cheung, MD, MPH<sup>†</sup>; Frances Mondor, MD\*

**CASE PRESENTATION**

A 48-year-old male was brought to a community emergency department (ED) after an in-flight collapse while on board a commercial aircraft. He had no complaints prior to boarding and had been on multiple previous commercial flights. He had no previous medical history and was on no regular medications. There was no history of alcohol or street drug abuse. Thirty minutes into the flight, he had a generalized tonic-clonic seizure and became comatose. The aircraft was immediately rerouted to get the patient to the nearest hospital. On ED arrival, the patient had a Glasgow Coma Scale score of 3, temperature of 36.6°C (97.8°F), blood pressure of 210/98 mm Hg, heart rate of 78 beats/min (normal sinus rhythm), respiratory rate of 28 breaths/min, oxygen saturation of 90% on 100% O<sub>2</sub>, and blood glucose of 11.0 mmol/L. On examination, there were no external signs of trauma. He had decreased breath sounds bilaterally, with normal heart sounds and no murmurs. His abdomen was soft and showed no evidence of distention. Both pupils were reactive to light but sluggish.

The patient was intubated in the ED for airway protection. A postintubation chest radiograph (Figure 1) was performed, which showed appropriate endotracheal tube depth and a large round opacity in the right lower lobe. An initial electrocardiogram showed a heart rate of 102 beats/min (sinus tachycardia) with no signs of right heart strain or ischemia. Initial bloodwork, including a complete blood count, electrolytes, renal function, liver function tests, lactate, international normalized ratio, and partial thromboplastin time, were all within normal limits, except for a white blood cell count of  $17.6 \times 10^9/L$  (normal  $< 11.0 \times 10^9/L$ ) and a troponin of 0.12 µg/L

(normal  $< 0.10 \mu\text{g/L}$ ). After intubation, the patient went immediately for a computed tomographic (CT) head scan (Figure 2), which was obtained approximately 2 hours after the in-flight seizure. As seen in the image, the CT head scan showed pneumocephalus with multiple locules of air in the brain parenchyma. Given these findings, the patient proceeded to have a CT chest scan (Figure 3) to identify a potential source for the gas emboli. The CT chest scan revealed a large  $8 \times 9 \times 10$  cm right lower lobe bronchogenic cyst with an air-fluid level. On the CT chest scan, the cyst was also thought to be communicating with a small bronchiole and the pulmonary vasculature.

With the diagnosis of cerebral gas emboli, the patient was immediately transferred to a tertiary care hospital for hyperbaric oxygen therapy. At the receiving hospital, to prevent further air emboli and isolate the right lung, bronchoscopy was performed; a right lung block was inserted, and the endotracheal tube was placed in the left main bronchus. A chest tube was placed directly into the bronchogenic cyst to allow for decompression. The patient was subsequently treated in the hyperbaric chamber. Despite aggressive treatment, the patient developed increasing cerebral edema with midbrain compression and bilateral uncal herniation. A postmortem wedge resection of the right lower lobe showed a bronchogenic cyst with empty fibrous-walled spaces and overlying reactive pleural changes indicative of previous air leaks.

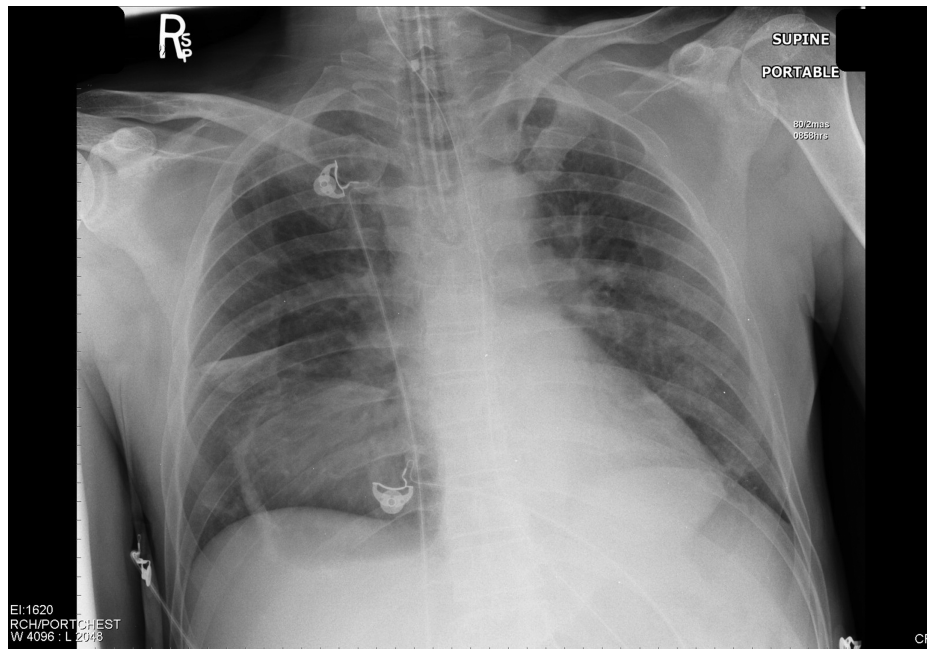
**LEARNING POINTS**

There are four main types of congenital lung cysts, including bronchogenic cysts, lobar emphysema,

From the \*Department of Emergency Medicine, Richmond Hospital, Richmond, BC, and the †Department of Emergency Medicine, Vancouver General Hospital, Vancouver, BC.

Correspondence to: Edward Mak, Richmond Hospital, 7000 Westminster Highway, Richmond, BC V6X 1A2; Edward.Mak@vch.ca.

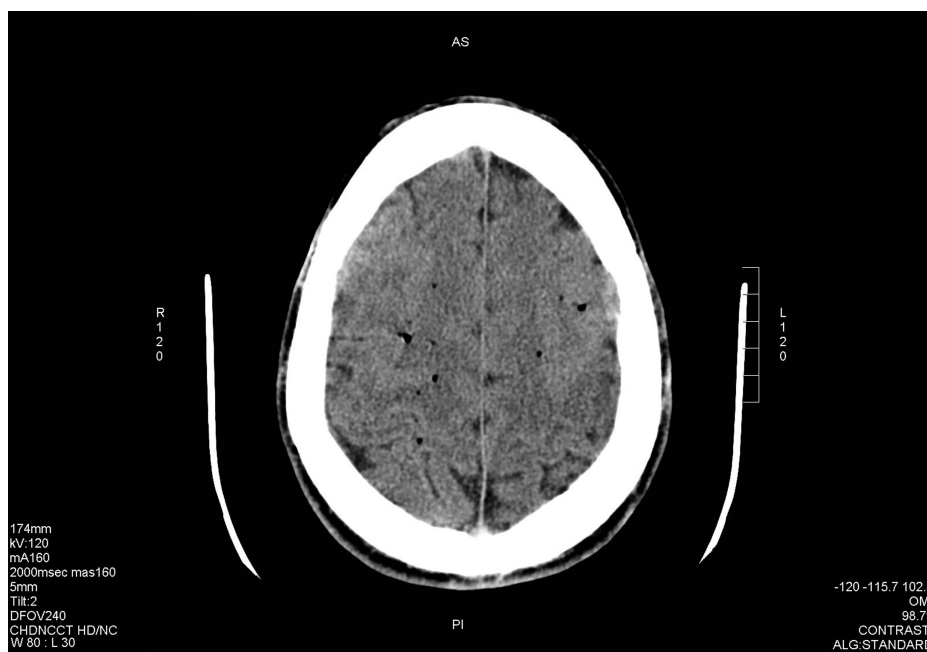
This article has been peer reviewed.



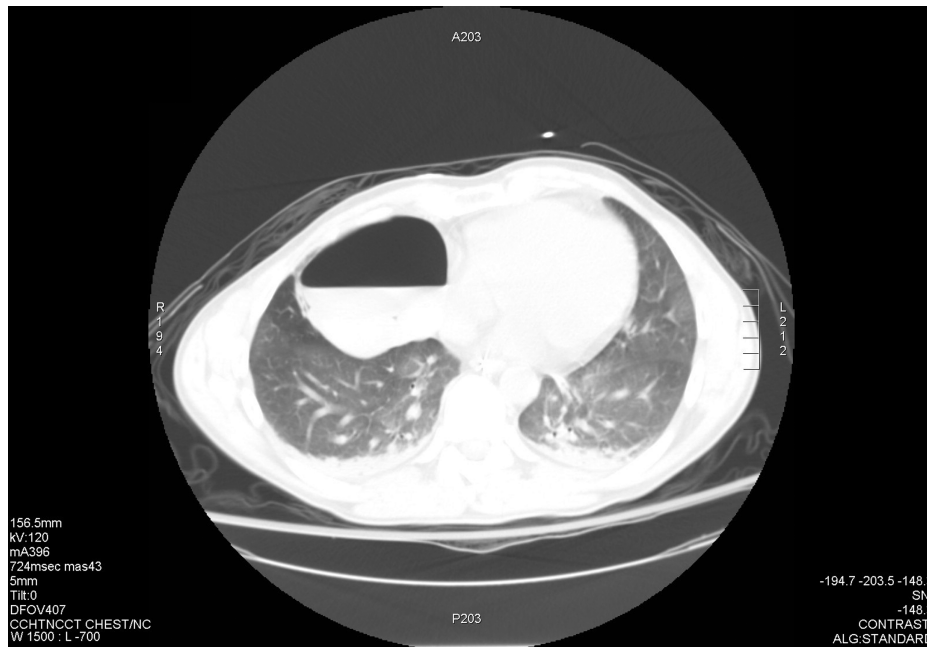
**Figure 1.** Standard radiographic image of the patient’s chest after intubation showing a large round opacity in the right lower lobe.

pulmonary sequestrations, and congenital cystic adenomatoid malformations.<sup>1</sup> The unilocular nature of the thin-walled pulmonary anomaly on this patient’s CT chest scan suggested a bronchogenic cyst, which was confirmed definitively by histopathology. Bronchogenic cysts are congenital malformations that arise from anomalous foregut budding<sup>2</sup> and are usually fluid filled. Although they are not associated with vascular malformations, they can communicate with

the tracheobronchial tree.<sup>3</sup> The prevalence of bronchogenic cysts is unknown as most are asymptomatic.<sup>4</sup> Serious complications may result, including acute respiratory decompensation, vascular compression, and air emboli.<sup>5-7</sup> As removal of symptomatic cysts may be associated with more complications than removal of asymptomatic cysts,<sup>5,6</sup> many authors have suggested that all incidental bronchogenic cysts be surgically removed.<sup>5,6,8</sup> A more recent review, where



**Figure 2.** Computed tomographic head scan of the patient at presentation showing multiple locules of intraparenchymal air.



**Figure 3.** Computed tomographic chest scan of the patient at presentation showing a large  $8 \times 9 \times 10$  cm cyst in the right lower lobe with an air-fluid level.

patients were followed for up to 22 years, showed that 45% of incidentally found asymptomatic bronchogenic cysts become symptomatic<sup>9</sup> and suggested that surgical excision and close follow-up of asymptomatic bronchogenic cysts are both viable options.

Our patient was diagnosed with an in-flight rupture of a bronchogenic cyst causing fatal cerebral gas emboli. This is an unusual complication of bronchogenic cysts, and there have only been five previous reports of similar cases.<sup>4,10–13</sup> In all of these cases, the pulmonary cyst occupied at least one-third to one-half of the involved hemithorax. Only two of these cases were confirmed to be bronchogenic cysts by pathology.<sup>4,11</sup> Our case is unique in that it is the first case report with clear documentation of intracerebral air on the CT head scan and gross pathology findings demonstrating reactive changes consistent with previous air leaks from the cyst.

On initial presentation, our patient was comatose and required intubation for airway protection. Although necessary, it is possible that positive pressure ventilation can increase the amount of air entering the cyst and increase subsequent systemic embolization. Once a pulmonary source of air emboli is identified, the involved lung should be isolated, and single-lung ventilation of the contralateral lung should be initiated. Airway pressures should be minimized prior to single-lung ventilation.

In-flight seizures are rare and occur in approximately 1 of every 650,000 airline passengers.<sup>14</sup> Cerebral

arterial gas embolism (CAGE) is a rare cause of in-flight seizures. Additional differential diagnoses to consider include other acute intracranial assaults, infection, metabolic derangements, drug-related etiologies, pseudoseizure, and other causes of syncope. During a flight, sleep disorders such as narcolepsy and periodic limb movements of sleep should also be considered. Our patient had multiple locules of air identified on CT head scan, confirming the diagnosis of CAGE. However, it is important to note that delayed CT imaging may miss intracerebral air and that small air emboli ( $< 1\text{--}2$  mm) may not be visualized.<sup>15</sup> Although there are no randomized, controlled trials, hyperbaric oxygen therapy is still considered first-line treatment for arterial gas embolism.<sup>16</sup> It decreases bubble size, decreases cerebral edema by promoting denitrogenation of brain tissue, improves oxygenation of hypoxic brain tissue,<sup>17</sup> and decreases inflammation.<sup>18</sup> In one case report, a comatose patient with CAGE secondary to a ruptured pulmonary cyst was treated with hyperbaric oxygen therapy 48 hours after presentation and made a full neurologic recovery.<sup>19</sup>

Although rare, CAGE should be considered when a patient develops sudden loss of consciousness or a seizure during a flight. Patients presenting after an in-flight seizure require a chest radiograph to assess for an intrapulmonary cyst.<sup>10</sup> Individuals with symptomatic pulmonary cysts require urgent medical management

and once stabilized, surgical excision of the cyst if possible. If no immediate therapy is indicated for an identified cyst, patients should be told to refrain from activities including air travel, mountain climbing, or diving.

**Competing interests:** None declared.

**Keywords:** bronchogenic cyst, cerebral gas emboli, commercial aircraft

## REFERENCES

1. Bailer PV, Tracy T Jr, Connors RH, et al. Congenital bronchopulmonary malformations. Diagnostic and therapeutic considerations. *J Thorac Cardiovasc Surg* 1990;99:597-602.
2. Berrocal T, Madrid C, Novo S, et al. Congenital anomalies of the tracheobronchial tree, lung, and mediastinum: embryology, radiology, and pathology. *Radiographics* 2004; 24:e173, doi:10.1148/rg.e17.
3. Ribet ME, Copin MC, Gosselin BH. Bronchogenic cysts of the lung. *Ann Thorac Surg* 1996;61:1636-40, doi:10.1016/0003-4975(96)00172-5.
4. Edwardson M, Wurth D, Lacy JM, et al. Cerebral air embolism resulting in fatal stroke in an airplane passenger with a pulmonary bronchogenic cyst. *Neurocrit Care* 2009;10: 218-21, doi:10.1007/s12028-008-9126-3.
5. Patel S, Meeker D, Biscotti C, et al. Presentation and management of bronchogenic cysts in the adult. *Chest* 1994; 106:79-85, doi:10.1378/chest.106.1.79.
6. St-Georges R, Deslauriers J, Durancea A, et al. Clinical spectrum of bronchogenic cysts of the mediastinum and lung in the adult. *Ann Thorac Surg* 1991;52:6-13, doi:10.1016/0003-4975(91)91409-O.
7. Bolton JW, Shahian DM. Asymptomatic bronchogenic cysts: what is the best management? *Ann Thorac Surg* 1992; 53:1134-7, doi:10.1016/0003-4975(92)90412-W.
8. Kosar A, Tezel C, Orki A, et al. Bronchogenic cysts of the lung: report of 29 cases. *Heart Lung Circ* 2009;18:214-8, doi:10.1016/j.hlc.2008.10.011.
9. Kirmani B, Firmani B, Sogliani F. Should asymptomatic bronchogenic cysts in adults be treated conservatively or with surgery? *Interact Cardiovasc Thorac Surg* 2010;11:649-59, doi:10.1510/icvts.2010.233114.
10. Arnaiz J, Marco de Lucas E, Piedra T, et al. In-flight seizures and fatal air embolism: the importance of a chest radiograph. *Arch Neurol* 2011;68:661-4, doi:10.1001/arch-neurol.2011.85.
11. Zaugg M, Kaplan V, Widmer U, et al. Fatal air embolism in an airplane passenger with a giant intrapulmonary bronchogenic cyst. *Am J Respir Crit Care Med* 1998;157:1686-9.
12. Almeida FA, Desouza BX, Meyer T, et al. Intrapulmonary bronchogenic cyst and cerebral gas embolism in an aircraft flight passenger. *Chest* 2006;130:575-7, doi:10.1378/chest.130.2.575.
13. Neidhart P, Suter PM. Pulmonary bulla and sudden death in a young aeroplane passenger. *Intensive Care Med* 1985;11:45-7, doi:10.1007/BF00256066.
14. Cummins RO, Schubach JA. Frequency and types of medical emergencies among commercial air travelers. *JAMA* 1989; 261:1295-9, doi:10.1001/jama.1989.03420090059031.
15. Dexter F, Hindman BJ. Recommendations for hyperbaric oxygen therapy of cerebral air embolism based on a mathematical model of bubble absorption. *Anesth Analg* 1997;84:1203-7.
16. Fukaya E, Hopf HW. HBO and gas embolism. *Neurol Res* 2007;29:142-5, doi:10.1179/016164107X174165.
17. Van Hulst RA, Klein J, Lachmann B. Gas embolism: pathophysiology and treatment. *Clin Physiol Funct Imaging* 2003;23:237-46, doi:10.1046/j.1475-097X.2003.00505.x.
18. Thom SR. Effects of hyperoxia on neutrophil adhesion. *Undersea Hyperb Med* 2004;31:123-31.
19. Closon M, Vivier E, Breynaert C, et al. Air embolism during an aircraft flight in a passenger with a pulmonary cyst: a favorable outcome with hyperbaric therapy. *Anesthesiology* 2004;101:539-42, doi:10.1097/00000542-200408000-00037.