

Case Report

Cerebral Air Embolism Following Endoscopic Stricture Dilatation

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Introduction

Direct communication between a source of air and the vasculature results in air embolism. The rate and the volume of air introduced into the circulation determine the effect of an air embolus. An air embolism is an uncommon but potentially catastrophic event. Many cases are subclinical with no adverse outcome. However, severe cases are characterized by acute vascular insufficiency of specific organs, such as the brain or the spinal cord leading on to hemodynamic collapse. Cerebral Air Embolism (CAE) is a critical clinical condition requiring a rapid diagnosis and therapeutic intervention. CAE is often associated with insertion, dysfunction, or disconnection of the central venous line [1]. Spontaneous or traumatic fistulas between air containing organs and the vascular system, surgery, and barotraumas are the other causes [2].

Case Presentation

A 40-year-old female patient presented with progressive dysphagia. She was diagnosed as achalasia cardia and underwent endoscopic balloon dilatation and per-oral endoscopic cardiomyotomy [POEM], following which, she developed esophageal perforation with intra-abdominal abscess. Emergency laparotomy was done with drainage of abscess and later esophageal reconstruction with colonic interposition. She was symptomatically better for a period of 4 months, but later on developed dysphagia, which progressively increased. Further evaluation revealed Stricture at the site of esophago-colonic anastomosis. Endoscopic dilation of the stricture was done. Immediately after the procedure, she became unresponsive with left focal seizures.

On clinical examination, her dolls eye movements were preserved but she had gaze preference to left. Pupils were equal and reactive to light. She had nystagmus to right, sluggish deep tendon reflexes and left extensor plantar response.

Abstract

Cerebral air embolism is a critical clinical condition requiring a rapid diagnosis and therapeutic intervention. We report a 40-year-old female patient with progressive dysphagia. Evaluation revealed esophageal stricture, following which endoscopic dilation was done. Immediately after the procedure, she became unresponsive with left focal seizures. Brain imaging was highly suggestive of air embolism in the right cerebral hemisphere with scattered acute infarcts. Along with this case report, we describe the pathophysiology, review of literature and treatment outcomes in regard to cerebral air embolism.

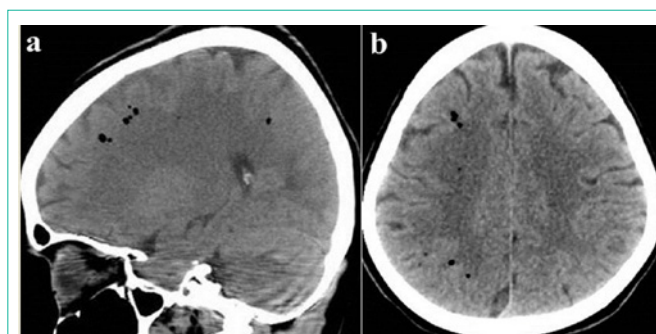


Figure 1: Non contrast axial [a] and sagittal [b] CT images show several foci of air within the cortical vessels in the right frontal and parietal region.

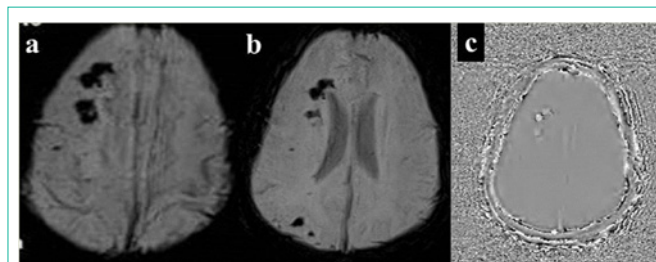


Figure 2: SWI [a], MIP [b], PHASE [c] images showing multiple low signal intensity foci, bright in phase image.

Non contrast CT scan (Figure: 1a,1b) showed several foci of air in the frontal and parietal region on the right side, suggestive of air embolism. Susceptibility Weighted (SW) images showed multiple punctuate low signal intensity foci in the right cerebral hemisphere in magnitude and MIP images (Figure: 2a,2b) and bright in phase images (Figure: 2c). There were small areas of diffusion restriction in the parietal cortex (Figure 3). Repeat CT after 24 hours showed infarct involving right MCA territory (Figure 4). However there was no evidence of residual pueumocephalus. Imaging features was

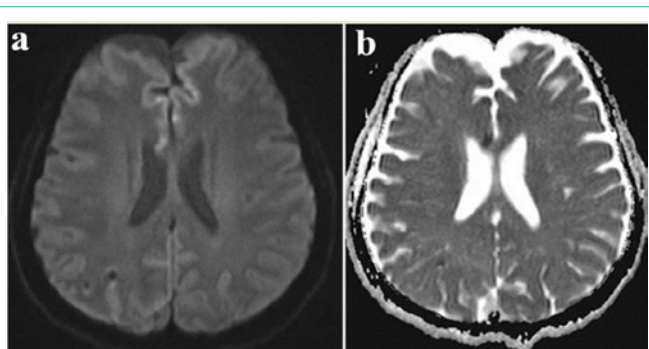


Figure 3: DW images [a] with ADC [b] showing small area of acute infarct in the cingulate gyrus.

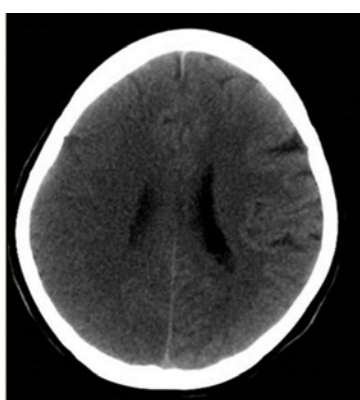


Figure 4: Repeat axial CT Scan showing acute infarct right fronto-parietal region with cortical hypo density, gyral effacement and loss of grey-white matter differentiation.

highly suggestive of air embolism in the right cerebral hemisphere with scattered acute infarcts. Although she was hemodynamically stable, the patient did not recover from the unconsciousness and finally succumbed to her illness.

Discussion

In CAE, diagnosis often depends on the clinical suspicion. The patient’s history combined with the clinical manifestations of the embolism due to the initial neurological insult and the direct temporal relation of procedures potential enough to cause air embolism are the most important diagnostic clues [3]. Computed tomography (CT) can detect air emboli early in the course of the disease. As reported in the literature, in CAE the right hemisphere seems to be more affected due to the course of the right brachiocephalic trunk which is the first main artery rising from the aortic trunk [3].

Air embolism is most commonly associated with ERCP, but can result from any endoscopic procedure (Table 1). Previous interventions or surgeries of the bile duct system, transhepatic portosystemic shunt, blunt or penetrating trauma to the liver, inflammation of the digestive system, post-surgical gastrointestinal fistula etc. are the reported risk factors for air embolism.

Interventional techniques resulting in air embolism include cholangioscopy, biliary sphincterotomy, metal stent placement, liver biopsy, insufflations of air with high pressure, excessive amount and/

Table 1: Reported Cases of Cerebral Air Embolism following Endoscopic procedures.

Case	Reference	Year	Age/Sex	Procedure	Outcome
1	Nayagam et al [6]	2004	57/M	ERCP	Dead
2	Mellado et al [7]	2005	52/F	Upper GI endoscopy	Dead
3	Rabe et al [23]	2006	87/M	ERCP	Survived
4	Stabile et al [24]	2006	65/M	ERCP	Dead
5	Tan et al [25]	2008	82/F	ERCP	Dead
6	McAree et al [8]	2008	69/M	Upper GI endoscopy	Dead
7	Rangappa et al [9]	2009	50/F	ERCP	Dead
8	Argüelles et al [10]	2009	82/M	ERCP	Dead
9	Laan et al [11]	2009	Not Available	Upper GI endoscopy	Survived
10	Bisceglia et al [12]	2009	78/M	ERCP	Dead
11	Goins et al [26]	2010	72/F	ERCP	Survived
12	van Boxel et al [27]	2010	82/M	ERCP	Survived
13	Lopez et al [13]	2010	61/F	Upper GI endoscopy	Died
14	Pandurangadu et al [28]	2010	71/M	Upper GI endoscopy	Survived
15	Maccarone et al [29]	2011	45/M	ERCP	Survived
16	Bechi et al [15]	2012	79/F	ERCP	Survived
17	Nern et al [30]	2012	58/F	ERCP	Dead
18	Efthymiou et al [31]	2012	62/F	ERCP	Survived
19	Donepudi et al [16]	2013	66/M	ERCP	Dead
20	Present Case	2016	40/F	Endoscopic stricture dilatation	Dead

or increased rate of air infusion, procedural site located higher than the level of the heart, and the use of nitrous oxide (N₂O)³.

The first documented case of air embolism in gastrointestinal endoscopy was described by Lowdon and Tidmore in 1988 [4]. In their report a 5-week-old infant who had biliary atresia due to a previous Kasai procedure died during endoscopy. Autopsy showed air in the right atrium and ventricle.

Christl et al [5] described the first incident of a cerebral air embolism following endoscopy in a patient with a duodenal ulcer and a duodenocaval fistula.

In 2004, Nayagam et al [6] reported a case of a 56-year-old man who died during ERCP due to cerebral ischemia caused by venous and arterial air embolism.

In 2005, Mellado et al [7] described a 52-year-old woman with ovarian cancer, who suffered from severe deterioration of her consciousness. A CT imaging performed revealed areas of multiple air embolism between the anterior and middle right cerebral arteries. Patient succumbed after 48hours.

In 2008, McAree et al [8] reported a cerebral air embolism in a subject being evaluated for metastatic adenocarcinoma of an unknown origin. Upper GI endoscopy showed erosive esophagitis. As the procedure was being completed, the patient became unresponsive and displayed neurological symptoms. An emergency cerebral CT confirmed air in the brain, specifically the right frontotemporal area.

In 2009, Rangappa et al [9] presented a case of a 50-year-old woman undergoing ERCP due to suspected choledocholithiasis, who suffered a fatal cerebral air embolism. After the procedure the patient was unresponsive with her gaze deviating to the right. CT scan of her brain showed air embolism in her right hemisphere with tonsillar herniation and diffused cerebral edema. Cause of death was brain edema due to paradoxical air embolism caused by injury of blood vessels at the site of sphincterotomy.

Argüelles et al [10] in 2009 reported a case of patient who died due to a cerebral artery air embolism during ERCP as he developed a severe ischemic brain injury.

In 2009, Laan et al [11] reported a patient, who underwent an upper endoscopy for evaluation of a gastric-mediastinal fistula after subtotal oesophagectomy and gastric tube reconstruction because of oesophageal cancer. During the procedure, cerebral gas emboli developed resulting in acute left-sided hemiparesis. Hyperbaric oxygen was administered, and the patient almost fully recovered.

Another similar case was reported by Bisceglia et al [12] a 78-year-old man underwent ERCP for recurrent cholangitis due to gallstones and developed cardiac arrest. Autopsy revealed massive pulmonary and cerebral air embolism.

In 2010, Lopez et al [13] presented a case of a 61-year-old woman who died of pneumocephalus and ischemic infarction of the right hemisphere. She underwent esophagogastroduodenoscopy due to hematemesis caused by esophageal varices. The patient developed hypotension and was unresponsive. Brain death was declared 24 hours post procedure.

Finsterer et al [14] in 2010 conducted a Medline search of 18 reports of 19 patients, in order to provide an overview of current knowledge of pathophysiology, diagnosis, management and prognosis of air embolism during ERCP. In 14 cases of air embolism after ERCP; 8 patients suffered cerebral air embolism and 6 of them ended fatally. The authors concluded that in the event if the subject does not wake up after the procedure, air embolism should be strongly taken into consideration and all of therapeutic measures provided.

In 2012, Bechi et al [15] described a case of a complete neurological recovery in a 79-year-old female patient who developed systemic air embolism during endoscopic sphincterotomy and gallstone removal. They provided only conservative treatment: 100% oxygen, head down position, lateral position and fluid resuscitation.

Pandurangadu et al [28] in 2012 reported a case of cerebral air embolism. A 71-year-old man received an outpatient upper GI endoscopy, which required an esophageal biopsy and ablation of duodenal arterio-venous malformations. Shortly after the procedure, he presented to the emergency room with neurological symptoms of sudden onset lethargy and left-sided weakness. CT scan of the brain showed multiple gas emboli in two areas, the right frontal lobe and fronto-parietal region.

Donepudi et al [16] in 2013 conducted a systematic review of the risk factors, clinical presentation and management of air embolism in gastrointestinal endoscopy. They observed 26 cases of air embolism as a complication of ERCP to date. Approximately half of these cases were cerebral air embolism.

Air on CT will have a very low density (near -1000HU) (HU – Hounsfield units) but fat with CT HU of -50 to -100 also appear completely black on routine brain windows. They can be differentiated based on CT attenuation values. CT can detect very small amounts of air and require only as low as 0.55ml to be detected [17].

On MRI, air will appear black on all sequences as it has few hydrogen protons and hence has low magnetic susceptibility. The signal loss is more evident on GRE (gradient echo) sequences, and the use of T2* GRE to facilitate detection of pneumocephalus has been described in a few articles [18,19]. In our case, air appeared as signal void in SWI (Susceptibility Weighted Imaging). SWI is a 3D high-spatial resolution gradient echo MRI sequence. After acquisition and post processing, the images obtained are magnitude phase, combined magnitude phase and minimum intensity projection images. We could not find any other article on SWI in cerebral air embolism.

Air embolism can be asymptomatic or cause mechanical obstruction leading to ischemia. Air bubbles are mostly lodged within the small arteries in the cortex. The entrapped air can also cause damage to the endothelium with breakdown of blood brain barrier, activating inflammatory response and leading to infarcts in cortical areas near the emboli [18] as in our subject.

Cerebral infarcts due to air are clearly visualized on DW (diffusion weighted) images and the air may rapidly disappear from the image [18]. There was no evidence of cerebral embolism in the repeat scan.

The cause of pneumocephalus in our case could be due to breach of esophageal mucosa. Venous blood from esophagus drains into sub mucous and periesophageal venous plexus then to azygos, hemiazygos, intercostal and bronchial veins and finally to SVC.

Cerebral venous air embolism could result due to the following mechanisms

- a) Retrograde movement of air into the jugular vein
- b) When the volume of embolus exceeds the capacity of pulmonary filter or
- c) If there is right to left intra cardiac shunting?

The lungs normally act as effective filter allowing only bubbles less than 22 micrometer to traverse the pulmonary vascular bed, the filtering capacity may be compromised by pulmonary barotrauma from positive pressure ventilation or when there is right to left shunting [18]. The most common cause of right to left shunts is PFO, ASD, AV shunt and intrapulmonary shunts. In some subjects, we cannot exclude the presence of small undetectable PFO or intrapulmonary shunt.

If air embolism is suspected, the following steps can have a significant impact on patient outcomes

- 1) Immediately stop the procedure.
- 2) Administer high flow 100% oxygen, which can reduce air bubbles expansion.
- 3) Place the patient in Trendelenburg (feet higher than the head) and left lateral decubitus position in order to minimize air migration to the brain and to force-out air from the right ventricular outflow tract, thereby increasing venous return.

4) Perform an emergency echocardiogram. If air is detected on the right side of the heart in the echocardiography, insert a central catheter.

5) Decompression with nasogastric suction.

6) Initiate high volume normal saline infusion.

7) If N₂O is being used, it must be discontinued because of its ability to rapidly diffuse into the trapped air bubbles, causing an additive effect on the embolism.

8) In the event of circulatory collapse, cardiopulmonary resuscitation (CPR) should be initiated in order to maintain the cardiac output. CPR may also serve to break large air bubbles into smaller ones and force air out of the right ventricle into the pulmonary vessels.

9) Start hyperbaric oxygen therapy as soon as the patient's condition allows it—subject to availability. Hyperbaric oxygenation therapy may reduce air bubble size, accelerate nitrogen reabsorption, and increase the oxygen content of arterial blood. This potentially reduces the ischemia.

10) Once the patient is hemodynamically stable, CT brain should be considered to confirm the diagnosis.

Cerebral air embolisms have a fatal prognosis. It can be prevented by using CO₂ for insufflation instead of air, because CO₂ can be easily absorbed [20,21]. Another option for subjects at risk is to use a precordial Doppler probe monitor during the procedure; which can quickly detect air within the heart and pulmonary vasculature before clinical symptoms may appear [22-31].

References

- Han SS, Kim SS, Hong HP, Lee SY, Lee SJ, Lee BK. Massive paradoxical air embolism in brain occurring after central venous catheterization: a case report. *J Korean Med Sci.* 2010; 25: 1536-1538.
- Nern C, Bellut D, Husain N, Pangalu A, Schwarz U, Valavanis A. Fatal cerebral venous air embolism during endoscopic retrograde cholangiopancreatography—case report and review of the literature. *Clin Neuroradiol.* 2012; 22: 371-374.
- Vachalová I, Ernst S, Vynogradova I, Wöhrmann S, Heckmann JG. Cerebral airembolism via port catheter and endoscopic retrograde cholangiopancreatography. *SpringerPlus.* 2013; 2: 477.
- Hauser G, Milosevic M, Zelić M, & Stimac D. Sudden Death After Endoscopic Retrograde Cholangiopancreatography (ERCP)—Case Report and Literature Review. *Medicine.* 2014; 93: 235.
- Christl SU, Scheppach W, Peters U, Kirchner T. Cerebral air embolism after gastroduodenoscopy: complication of a duodenocaval fistula. *Gastrointest Endosc.* 1994; 40: 376-378.
- Nayagam J, Ho KM, Liang J. Fatal systemic air embolism during endoscopic retrograde cholangio-pancreatography. *Anaesth Intensive Care.* 2004; 32: 260-264.
- Mellado TP, Constanzo PF, Miguel PJF, et al. Ischemic brain infarction after an air embolism. *Case report. Rev Med Chil.* 2005; 133: 453-456.
- McAree BJ, Gilliland R, Campbell DM, Lucas JW, Dickey W. Cerebral air embolism complicating esophagogastroduodenoscopy (EGD) *Endoscopy.* 2008; 40: 191-192.
- Rangappa P, Uhde B, Byard RW, et al. Fatal cerebral arterial gas embolism after endoscopic retrograde cholangiopancreatography. *Indian J Crit Care Med.* 2009; 13: 108-112.
- Argüelles García B, García Blanco A, Meilán Martínez A, et al. Cerebral artery air embolism secondary to endoscopic retrograde cholangiopancreatography. *GastroenterolHepatol.* 2009; 32: 614-617.
- ter Laan M, Totte E, van Hulst RA, et al. Cerebral gas embolism due to upper gastrointestinal endoscopy. *Eur J Gastroenterol Hepatol.* 2009; 21: 833-835.
- Bisceglia M, Simeone A, Forlano R, et al. Fatal venous air embolism during endoscopic retrograde cholangiopancreatography. *Adv Anat Pathol.* 2009; 16: 255-262.
- Lopez JC, Perez X, Esteve F. Cerebral air embolism during upper endoscopy. *Endoscopy* 2010; 42: 41.
- Finsterer J, Stöllberger C, Bastovansky A. Cardiac and cerebral air embolism from endoscopicretrograde cholangiopancreatography. *Eur J Gastroenterol Hepatol.* 2010; 22: 1157-1162.
- Bechi A, Nucera MP, Olivotto I, et al. Complete neurological recovery after systemic air embolism during endoscopic retrograde cholangiopancreatography. *MinervaAnesthesiol.* 2012; 78: 622-625.
- Donepudi S, Chavalitdhamrong D, Pu L, Draganov PV. Air embolism complicating gastrointestinal endoscopy: A systematic review. *World Journal of GastrointestinalEndoscopy.* 2013; 5: 359-365.
- Dabdoub CB, Salas G, N. Silveira Ed, Dabdoub CF. Review of the management of pneumocephalus. *Surg Neurol Int.* 2015; 6: 155.
- Yoko Kaichi, Shingo Kakeda, Yukunori Korogi, et al. "Changes over Time in Intracranial Air in Patients with Cerebral Air Embolism: Radiological Study in Two Cases," *Case Reports in Neurological Medicine.* 2015.
- Brar R, Prasad A, Sharma T, Khosla VK. Pneumocephalus on MRI: Use of gradient imaging. *Indian J Neurotrauma.* 2011; 8: 59-60.
- Romberg C. Systemic air embolism after ERCP: a case report and review of the literature (with video) *Gastrointest Endosc.* 2009; 70: 1043-1045.
- Dellon ES, Hawk JS, Grimm IS, Shaheen NJ. The use of carbon dioxide for insufflation during GI endoscopy: a systematic review. *Gastrointest Endosc.* 2009; 69: 843-849.
- Siddiqui J, Jaffe PE, Aziz K, Forouhar F, Sheppard R, Covault J, Bonkovsky HL. Fatal air and bile embolism after percutaneous liver biopsy and ERCP. *GastrointestEndosc.* 2005; 61: 153-157.
- Rabe C, Balta Z, Wüllner U, Heller J, Hammerstingl C, Tiemann K, Sommer T, Schepke M, Fischer HP, Sauerbruch T. Biliary metal stents and air embolism: a note of caution. *Endoscopy.* 2006; 38: 648-650.
- Stabile L, Cigada M, Stillitano D, Morandi E, Zaffroni M, Rossi G, Lapichino G. Fatal cerebral air embolism after endoscopic retrograde cholangiopancreatography. *ActaAnaesthesiol Scand.* 2006; 50: 648-649.
- Tan BK, Saunier CF, Cotton F, Gueugniaud PY, Piriou V. [Thoracoabdominal CT scan: a useful tool for the diagnosis of air embolism during an endoscopic retrograde cholangiopancreatography]. *Ann Fr Anesth Reanim.* 2008; 27: 240-243.
- Goins KM, May JM, Hucklenbruch C, Littlewood KE, Groves DS. Unexpected cardiovascular collapse from massive air embolism during endoscopic retrograde cholangiopancreatography. *Acta Anaesthesiol Scand.* 2010; 54: 385-388.
- van Boxel GI, Hommers CE, Dash I, Goodman AJ, Green J, Orme RM. Myocardial and cerebral infarction due to massive air embolism following endoscopic retrograde cholangiopancreatography (ERCP). *Endoscopy.* 2010; 42: 80-81.
- Pandurangadu AV, Paul JAP, Barawi M, Irvin CB: A case report of cerebral air embolism after esophagogastroduodenoscopy: diagnosis and management in the emergency department. *J Emerg Med.* 2012; 43: 976-979.
- Maccarone G, Shakoort T, Ellis B. Air embolism after percutaneous transhepatic biliary drainage and subsequent endoscopic retrograde cholangiopancreatography (ERCP). *Endoscopy.* 2011; 399.
- Nern C, Bellut D, Husain N, Pangalu A, Schwarz U, Valavanis A.

Fatal cerebral venous air embolism during endoscopic retrograde cholangiopancreatography-case report and review of the literature. Clin Neuroradiol. 2012; 22: 371–374.

31. Efthymiou M, Raftopoulos S, Antonio Chirinos J, May GR. Air embolism complicated by left hemiparesis after direct cholangioscopy with an intraductal balloon anchoring system. Gastrointest Endosc. 2012; 75: 221–223.