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CASE REPORT

Fatal air embolism during intestinal endoscopy in Kasai portoenterostomy for biliary atresia: A case report

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Abstract

BACKGROUND

Air embolism (AE) is a rare but potentially fatal complication of intestinal endoscopy (IE).

CASE SUMMARY

Herein, we report the case of an 18-year-old woman who underwent a successful Kasai portoenterostomy (KPE) for biliary atresia but died of AE during intraoperative IE for stone removal at the portoenterostomy site. Our review of the English literature identified only four similar cases of fatal AE during IE in patients undergoing KPE. The common clinical setting in the five patients, including our case, was high-pressure air insufflation into the blind closed afferent loop of the KPE to secure visibility. We hypothesize that the highly pressurized air injected into the closed loop entered the bile canaliculi—previously opened by KPE for bile drainage—passed through the tiny, microscopic pores of the fenestrated liver sinusoid endothelial cells, and finally entered the bloodstream with ease, resulting in fatal AE.

CONCLUSION
Meticulous performance of IE, especially on the KPE blind loop, is warranted owing to the risk of AE.

Key Words: Air embolism; Intestinal endoscopy; Biliary atresia; Kasai portoenterostomy; Fenestrated liver sinusoidal endothelial cell; Case report

Core Tip: Low-pressure CO_2 insufflation instead of high-pressure air insufflation is more suitable during intestinal endoscopy in patients with Kasai portoenterostomy to prevent fatal air embolism.

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INTRODUCTION

Air embolism (AE) during intestinal endoscopy (IE) is rare. Small AEs, if absorbed spontaneously, are subclinical with no adverse outcomes. However, large amounts of air infused into the blood circulation may be fatal due to AE. The first case of fatal AE during IE was reported in 1988 in an infant who had previously undergone Kasai portoenterostomy (KPE) for biliary atresia (BA)[1]. To date, three more cases of KPE with AE, with two of them being fatal, have been reported under a similar endoscopic study setting as in the first case[2-4]. We recently encountered a case of fatal AE during an IE examination in an 18-year-old woman with a history of successful KPE. Unfortunately, a massive AE during intraoperative IE resulted in her sudden and unexpected death.

The purpose of this report is to share the experience of contending with a fatal AE occurring during IE in patients who have undergone KPE and propose a hypothesis regarding the mechanism by which massive AE may occur in such patients.

CASE PRESENTATION

Chief complaints

The patient presented with jaundice as the chief complaint.

History of present illness

An 18-year-old woman was transferred to our hospital after 7 days of admission at a local hospital for acute cholangitis and hepatic duct stone. She was originally diagnosed with BA at 43 days of age and underwent KPE, which was performed by the corresponding author. After KPE, complete disappearance of the jaundice and gradual improvement in the hepatic fibrosis score with eventual normalization were noted. She was followed up at half-year intervals at the outpatient clinic of our hospital with medication (ursodeoxycholic acid, 3-400 mg/day), during which, cholangitis was not noted. However, an abdominal computed tomography performed at the local hospital before transfer showed a hyperdense lesion in the hepatic hilum, suggesting bile sludge or stone (Figure 1A).

History of past illness

Agitated saline transthoracic contrast echocardiography for intrapulmonary or intracardiac shunt showed the injected microbubbles filling the left atrium and ventricle of the heart within 7-8 heartbeats after intravenous injection of agitated saline, suggesting a minimal non-pathological intrapulmonary shunt. During the follow-up for BA at our hospital, serial imaging studies, including abdominal ultrasonography and magnetic resonance imaging (MRI), revealed progressive atrophic changes in the right lobe of the liver with compensatory hypertrophy of the left hepatic lobe; additionally, the formation of multiple tiny intrahepatic cysts without abnormal shadows suggestive of bile plugs or stone formation were noted.

Imaging examinations

Following admission to our hospital for suspected acute cholangitis with a stone in the hepatic hilum, abdominal MRI revealed a T2 low-signal intensity lesion at the KPE site (Figure 1B), suspected to be a stone or sludge lump. KPE site stones or sludge were inferred to be the likely causes of the cholangitis.

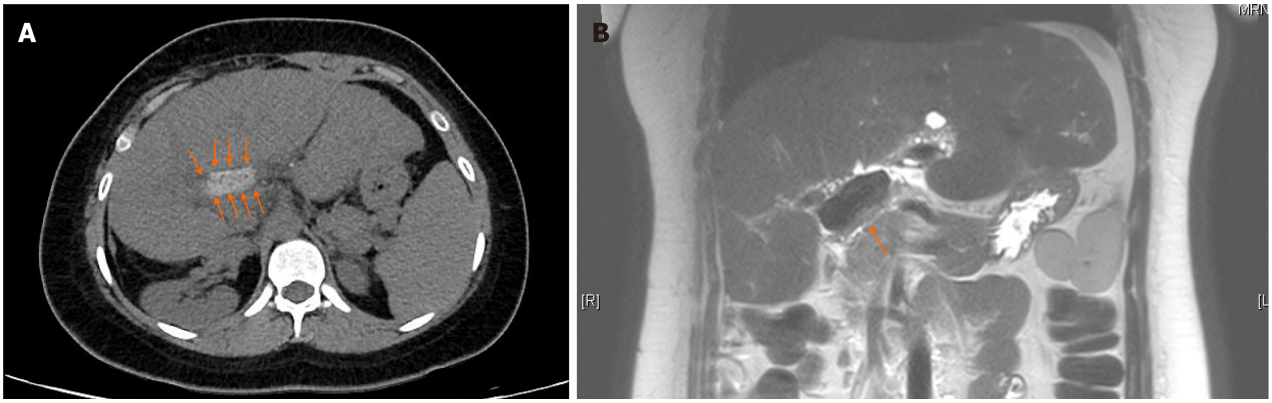

Figure 1 A stone at Kasai portoenterostomy sites in computed tomography and magnetic resonance imaging. A: The axial images of the abdominal computed tomography scan, taken in the local hospital before transfer, show a hyperdense lesion (arrows) at the hepatic hilum suggesting bile sludge or stone; B: The coronal image of the abdominal magnetic resonance imaging taken at our hospital after transfer shows a T2 low-signal intensity lesion (arrow) at the Kasai portoenterostomy site.

FINAL DIAGNOSIS

The final diagnosis was biliary stone at KPE site.

TREATMENT

Following treatment for cholangitis with broad-spectrum antibiotics, an attempt at percutaneous transhepatic biliary drainage for removal of the stone failed owing to lack of ductal dilatation on sonography. Therefore, we decided to remove the stone from the KPE *via* an intraoperative endoscopy. After adhesiolysis of the subhepatic space, the afferent blind jejunal loop of the KPE was identified, and a small jejunotomy was performed. A flexible intestinal endoscope (GIF-Q260, 9.2 mm; Olympus, Tokyo, Japan) was inserted into the jejunotomy site and slowly advanced to the blind end of the KPE site while insufflating air. During the procedure, no leak of insufflated air from the jejunotomy site was observed as the diameter of the endoscope fit the size of the jejunotomy opening perfectly. IE revealed a large, thick, yellow bile sludge-like lump hanging from the KPE site, which was easily broken during endoscopic removal (Figure 2).

Approximately 3 minutes after starting the endoscopic procedure, end-tidal CO_2 was suddenly undetectable, and the electroencephalography wave on the anesthetic depth monitor flattened. The patient's heart rate changed from a normal sinus rhythm to severe bradycardia and asystole, the blood pressure suddenly dropped, and oxygen saturation was not measured. Rapid cardiopulmonary resuscitation, including chest compressions and repeated administration of epinephrine and vasopressin, was initiated. However, immediately after two cycles of cardiac compression and epinephrine administration, uncontrollable frothy pink bleeding from the endotracheal tube was noted. As the patient had been previously healthy, no intraoperative difficulties such as bleeding had been anticipated. We had inserted only two large-bore peripheral intravenous catheters for anesthesia. Anesthesiologists attempted to insert a central venous catheter through the internal jugular vein for proper resuscitation; however, no vessels were detected on ultrasonographic examination. Therefore, femoral venous cannulation was performed, and air bubbles were noted in the blood withdrawn from the venous cannula. Intraoperative transesophageal echocardiography confirmed the presence of a large amount of air in all four heart chambers. Simple chest radiography also showed shadows of entrapped air in the cardiac chambers (Figure 3). Subsequently, venoarterial extracorporeal membrane oxygenation was successfully applied through the right femoral vessels; however, this took approximately 30 minutes because of the collapsed vessels and an unstable surgical field due to continuous chest compression.

OUTCOME AND FOLLOW-UP

Despite successful extracorporeal membrane oxygenation, progression of the patient's condition to multiple organ failure following no restoration of vital signs led us to declare the patient dead 5 hours after cardiac arrest. Although recommended, autopsy was not performed on account of her parents' refusal (Table 1).

DISCUSSION

AE during IE is rare but fatal. In a survey of the members of the American Society for Gastrointestinal Endoscopy in 1976 [5], no episodes of AE were found in 211410 IE procedures. According to a systematic review of the PubMed database on

Table 1 Timeline of patient's course

Timeline	Patient's course
HOD 1	Admission for jaundice and stone at KPE site
HOD 6	MRCP
HOD 11	Attempted PTBD, but the procedure failed due to the patient's uncooperativeness
HOD 16, 08:40 AM	Operation starts
12:00 PM	Intraoperative endoscopy insertion
12:03 PM	Arrhythmia, hypotension, bradycardia
12:10 PM	Asystole, CPR start
13:07 PM	ECMO insertion, compression stop
15:15 PM	Operation end, the patient was sent to ICU
17:10 PM	The patient expired.

KPE: Kasai portoenterostomy; HOD: Hospital day; MRCP: Magnetic resonance cholangiopancreatography; PTBD: Percutaneous transhepatic biliary drainage; CPR: Cardiopulmonary resuscitation; ECMO: Extracorporeal membrane oxygenation; ICU: Intensive care unit.

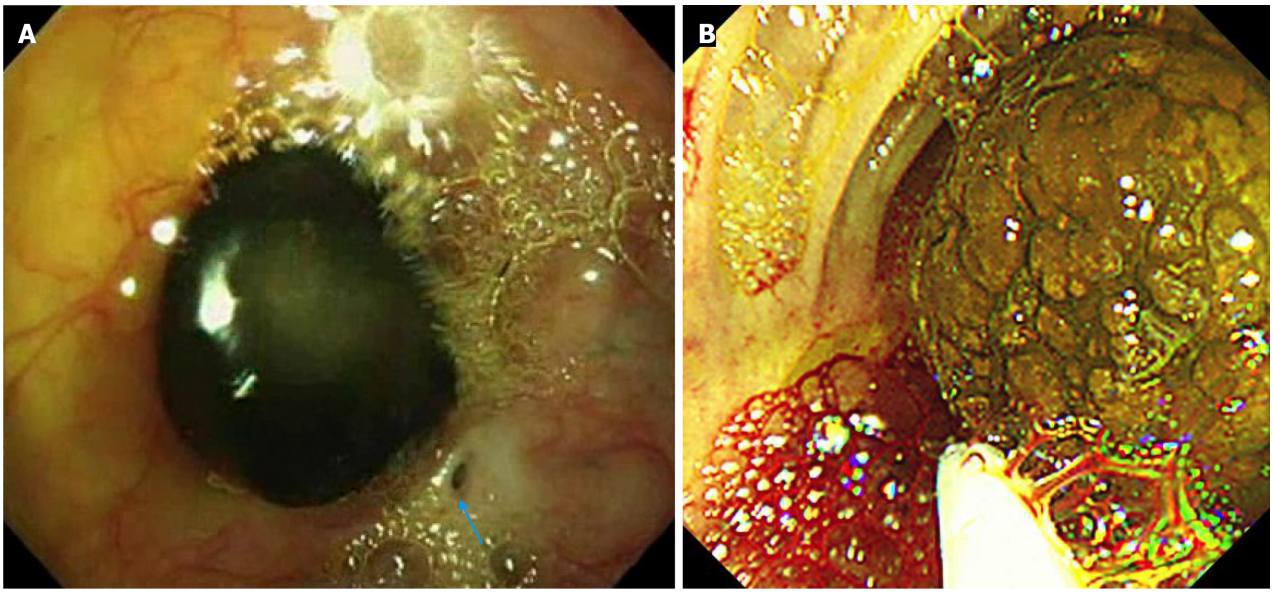

Figure 2 Intraoperative endoscopic view of stone. A: Intraoperative endoscopic findings in an unpublished previous case of a Kasai portoenterostomy (KPE) site stone, experienced 14 years ago, showing a black pigmented stone hanging like a stalactite from the KPE site and a tiny opening of a bile ductule (arrow). Endoscopic removal of the stone resolved the cholangitis and prevented further development of cholangitis in this case; B: Intraoperative endoscopic findings in the current case showing a large thick bile sludge-like lump hanging from the KPE site, which was easily broken during endoscopic removal. Three minutes after starting the endoscope examination, a sudden fatal air embolism occurred, and the patient died.

reported AE, 41 cases of AE during IE were identified in the English literature up to 2013[6]. The first case of AE during IE was reported in 1988 by Lowdon and Tidmore[1], involving a 4-month-old infant with a history of KPE for BA who died from fatal AE during intraoperative endoscopic exploration of the afferent loop of KPE. Since then, three additional cases of fatal AE in patients with KPE have been reported in the English literature (Table 2)[1-4]. Several conditions must be fulfilled for AE to occur during an IE. For air embolization to occur, air must pass through the gap in the physical barrier between the intestine and blood vessels. Moreover, there must be an interruption in the air-inflated intestine or bile duct barrier[3]. AE is more frequently observed during endoscopic exploration of the biliary system, such as endoscopic retrograde cholangiopancreatography, than during other intestinal endoscopic procedures[6]. The microscopic anatomy of the sinusoidal structure of the liver is critical in the occurrence of AE. Liver sinusoids comprise liver sinusoidal endothelial cells (LSECs) and are distinct from other capillaries in the body because of their open pores characterized by the absence of both a diaphragm and basal lamina beneath the endothelial layer (Figure 4). LSEC crucially regulates the exchange of macromolecules, solutes, and fluids between blood and surrounding tissues, such as bile canaliculi, through these fenestrae[7]. When high-pressure air is insufflated into a closed blind loop during IE in patients with KPE, air enters the bile canaliculi, passes through these tiny fenestrae in the LSEC, and easily enters the

Table 2 Summary of common clinical settings in the cases of fatal air embolism in patients with Kasai portoenterostomy for biliary atresia

Ref.	Age	Indication	Endoscopic entry	Air insufflation to closed loop	Time to arrest (minutes)	Paradoxical embolism	Result
Lowdon and Tidmore [1], 1988	4 months	Cholangitis	Enterotomy of A-loop	Yes	5	Yes	Died (autopsy)
Desmond and MacMahon[2], 1990[2]	10 years	Stone of KPE site	Enterotomy of A-loop	Yes	10	Yes	Died (autopsy)
Park *et al*[3], 2010	17 years	Intestinal bleeding	Enterotomy of A-loop	Yes	Immediately	Yes	Survived
Pujari *et al*[4], 2012	5 years	Intestinal bleeding	Enterotomy of A-loop	Yes	1	Yes	Died (no autopsy)
2023 (present case)	18 years	Stone at KPE site	Enterotomy of A-loop	Yes	3	Yes	Died (no autopsy)

Age: Patient's age when endoscopy was performed; A-loop: Afferent loop; KPE: Kasai portoenterostomy.

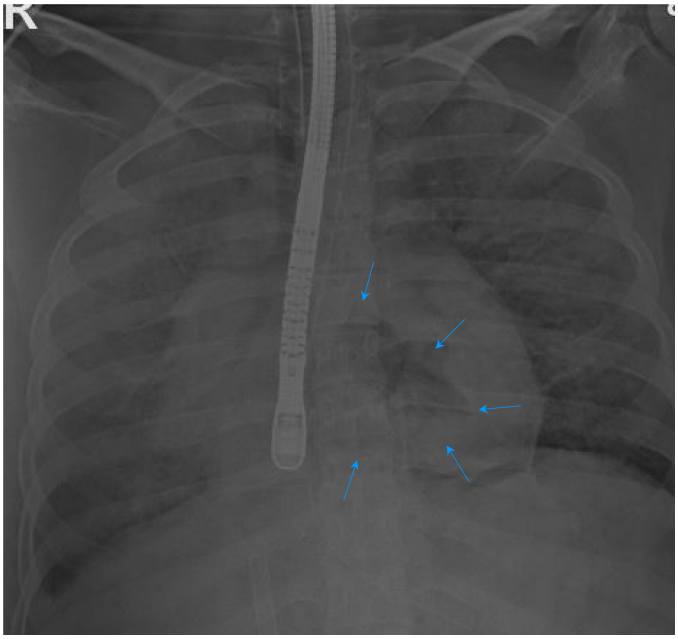

Figure 3 Air embolism in cardiac chamber (arrow) confirmed with chest X-ray taken at the operating room.

circulating bloodstream of the venous system, potentially leading to fatal AE (Figure 5). Next, air in the venous system can enter the systemic circulation through several mechanisms, including intracardiac or intrapulmonary shunting, or incomplete filtration of the intrapulmonary vessels, such as a paradoxical shunt[8,9]. The patient in this case also had a minimal intrapulmonary shunt, which could have acted as a pathway for air in the venous system to enter systemic circulation. In patients with a history of KPE, IE to explore the biliary tract can only be performed through the afferent blind loop of the Roux-en-Y limb. This approach can result in higher-pressure air insufflation compared to other endoscopic procedures, which may easily lead to AE during IE[10].

Therefore, to prevent fatal AE we recommend using low-pressure CO_2 insufflation instead of air insufflation through a blind afferent loop during IE in patients with hepatobiliary disease, such as those who have undergone biliary duct surgery, including biliary-enteric anastomosis, especially in patients with KPE.

CONCLUSION

AE during IE is rare but can be life-threatening. In this case, we hypothesized how severe AEs may develop in patients with KPE during IE. We recommend using low-pressure CO_2 insufflation instead of high-pressure air insufflation during IE in patients with KPE to prevent fatal AE.

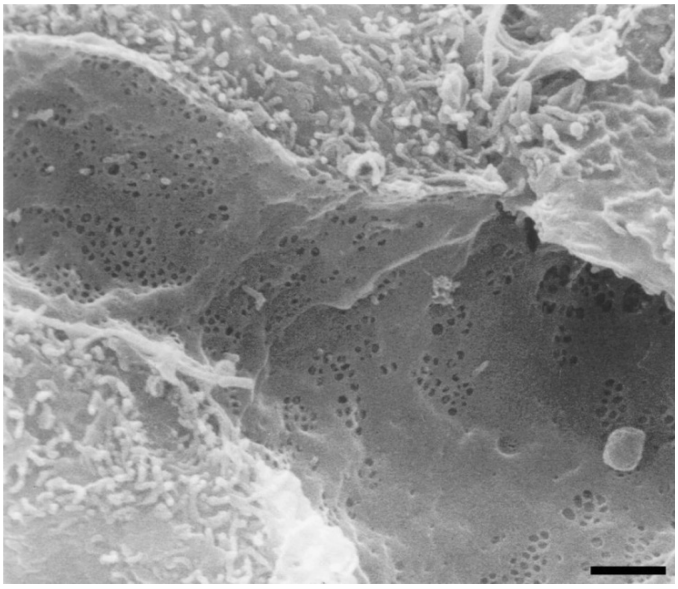

Figure 4 Low magnification scanning electron micrograph of the sinusoidal endothelium from a rat liver showing the fenestrated wall. Shown is the clustering of fenestrae in sieve plates. Scale bar: 1 μm. Citation: Braet F, Wisse E. Structural and functional aspects of liver sinusoidal endothelial cell fenestrae: a review. *Comp Hepatol* 2002; 1(1): 1. Copyright ©The Author(s) 2019. Published by BioMed Central Ltd[7].

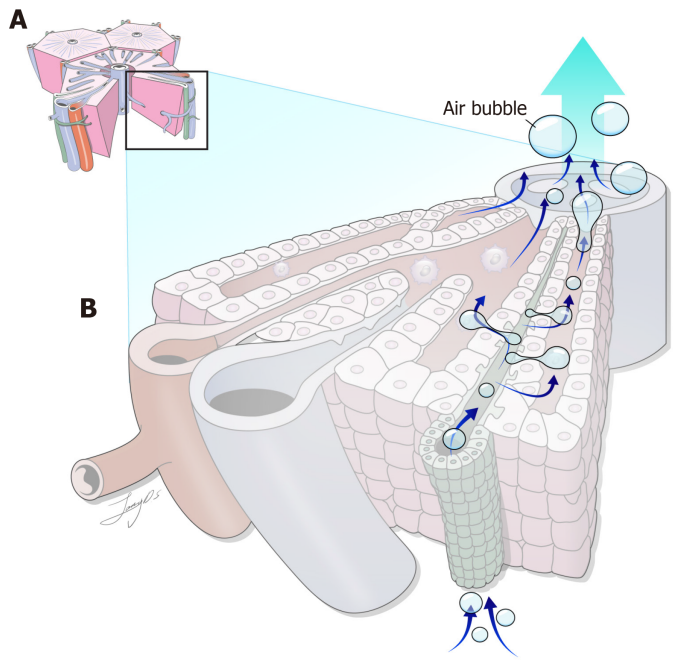

Figure 5 When high-pressure air is insufflated into a closed blind loop during intestinal endoscopy, as in patients with Kasai portoenterostomy, air may enter the bile canaliculi and pass into the bloodstream through these tiny pores of liver sinusoid endothelial cells, potentially leading to fatal air embolism.

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FOOTNOTES

Author contributions: Shin SY and Han SJ designed and performed the literature review and wrote the case report; Yeon HJ, Lee SO, Lee JR, Leem G and Han SJ were involved with the surgery and managed the patient.

Informed consent statement: The patient and her parents provided written informed consent prior to the surgery.

Conflict-of-interest statement: The authors declare no conflicts of interest related to this case report.

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