

Anesthetic management for a patient with bronchobiliary fistula after pancreaticoduodenectomy

A case report

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Abstract

Rationale: Perioperative management of patients with bronchobiliary fistula (BBF) is an anesthetic challenge because they typically exhibit poor lung function preoperatively, require meticulous lung isolation intraoperatively and need postoperative respiratory support.

Patient concerns: A 44-year-old man with a past surgical history of pancreaticoduodenectomy presented fluctuating fever, jaundice, dyspnea and yellowish sputum. Despite intravenous antibiotic treatment and repeated percutaneous drainage, patient showed gradual deterioration with hypoxemia, and uncontrolled pneumonia.

Diagnoses: The patient was diagnosed with BBF based on the clinical manifestation such as biloptysis with pneumonia, and imaging studies.

Interventions: Resection of the fistula and lobectomy was performed under general anesthesia. Avoidance of positive pressure ventilation before lung isolation and precise lung isolation are essential for patients with BBF to protect the unaffected lung. Therefore, rapid sequence induction was performed. Left-sided double-lumen tube was inserted for lung isolation and position of the tube was confirmed by visualization with fiberoptic bronchoscopy. Bile-stained secretion was repeatedly suctioned in trachea and both bronchi during surgery.

Outcomes: In spite of decrease in SpO₂ with institution of one-lung ventilation, the patient's oxygenation was gradually improved as surgery progressed without hemodynamic instability. At the end of surgery, the double-lumen tube was replaced with a single-lumen endotracheal tube for postoperative mechanical ventilation.

Lessons: Absolute lung isolation using double-lumen tube for one-lung ventilation and bronchial toilet during surgery and replacement of single-lumen tube for postoperative respiratory support at the end of surgery are effective to improve oxygenation in patients with BBF.

Abbreviations: ABGA = arterial blood gas analysis, ABP = arterial blood pressure, BBF = bronchobiliary fistula, CT = computed tomography, CVP = central venous pressure, DLT = double-lumen tube, ERBD = endoscopic retrograde biliary drainage, FOB = fiberoptic bronchoscopy, OLV = one-lung ventilation, PIP = peak inspiratory pressure, PPV = positive-pressure ventilation, PTBD = percutaneous transhepatic biliary drainage, TLV = two-lung ventilation.

Keywords: biliary drainage, bronchobiliary fistula, lobectomy, one-lung ventilation, pancreaticoduodenectomy

1. Introduction

Bronchobiliary fistula (BBF) is a rare disorder characterized by pathological communication between the biliary tract and

bronchial tree. It develops from numerous etiologies including congenital diseases, trauma, infection, tumor invasion, and bile duct obstruction. It may also occur secondary to various therapeutic interventions.^[1,2] Because alkaline bile irritates and erodes liver, diaphragm, lung parenchyma and bronchi, patients with BBF present with biloptysis, acute dyspnea and broncho-pneumonia.^[1] BBF may heal if subdiaphragmatic abscess is adequately drained and lesions in the bile ducts are corrected. Surgical resection of the fistula and involved lung is necessary if conservative treatment has failed or when there are complications from underlying disease.^[3] However, optimal management of BBF remains controversial depending on the disease severity and extent, and is associated with significant morbidity and mortality rates.^[1]

Patients with BBF are an anesthetic challenge because they exhibit poor lung function preoperatively, require absolute lung isolation intraoperatively and need postoperative respiratory support. However, the optimal anesthetic management in these patients is rare in the literature. We describe the anesthetic management of a male patient who underwent resection of fistula

Editor: N/A.

The authors declare no funding and conflict of interest.

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Medicine (2019) 98:19(e15694)

Received: 20 December 2018 / Received in final form: 10 April 2019 / Accepted: 22 April 2019

<http://dx.doi.org/10.1097/MD.00000000000015694>

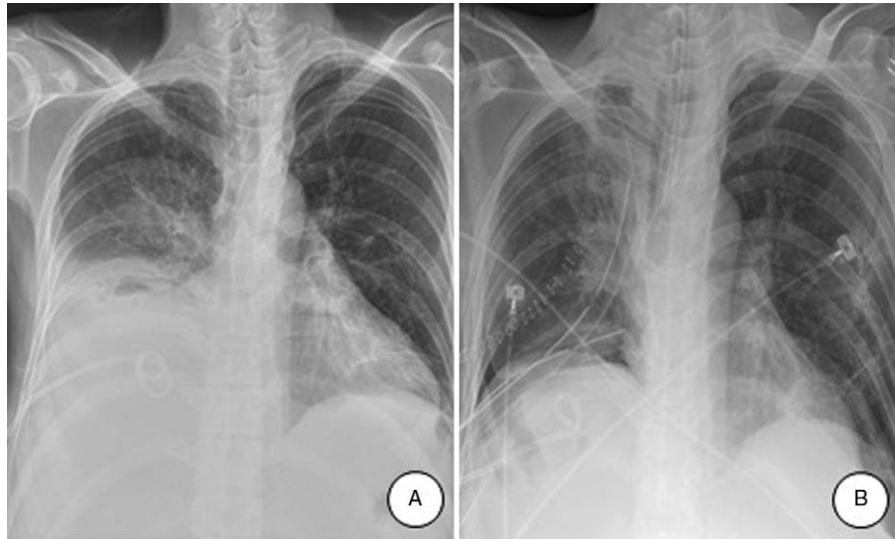


Figure 1. (A) Preoperative chest X-ray reveals consolidation of the right lower lung and bilateral pleural effusion. (B) Postoperative chest X-ray reveals drainage of right pleural effusion.

and bilobectomy of right lung using one-lung ventilation (OLV) for treatment of BBF and pneumonia.

2. Case report

A 44-year-old man (47.8 kg, 168 cm) was referred to thoracic department due to bronchobiliary fistula with pneumonia unresponsive to percutaneous biliary drainage and antibiotic treatment. He had a past surgical history of pancreaticoduodenectomy (Whipple operation) for carcinoma of the ampulla of Vater; subsequently, he had undergone transarterial embolization for pseudoaneurysm rupture of the gastroduodenal artery 4 months ago. His medical history was uneventful until dilation of the bile duct and small-sized multiple bilomas were detected in hepatic segment VIII 1 months ago. His initial symptoms were poor oral intake, fluctuating fever and jaundice. Despite

intravenous antibiotic treatment and repeated drainage endoscopic (ERBD) or percutaneous (PTBD) drainage, recurrent biliary stent obstruction resulted in acute dyspnea with yellowish sputum 1 week ago prior to the beginning of this case. The sputum was positive for bilirubin using a urine dipstick. Institutional review board of Yeungnam University Hospital approved this case report and waived informed consent due to death of patient and anonymity in publication of the case.

Preoperative chest X-ray demonstrated progressive consolidation and pleural effusion in the right lung (Fig. 1). Abdominal computed tomography (CT) revealed dilated bile duct and bilomas of various sizes in liver. Chest CT revealed subdiaphragmatic abscess, pneumonic consolidation with necrotizing change in the right lower lung and bilateral pleural effusion (Fig. 2). A tubogram revealed a fistulous communication between the dilated intrahepatic bile duct and right lower lung. Fiberoptic

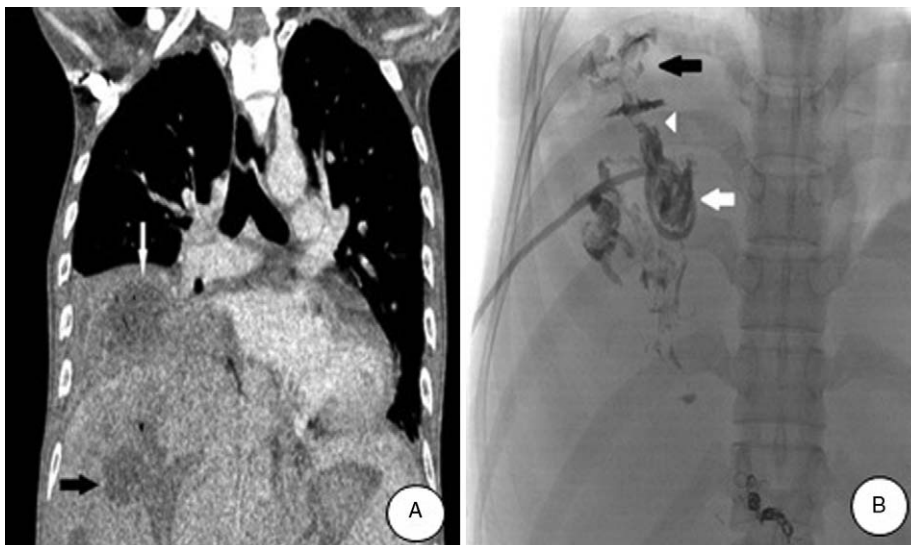


Figure 2. (A) A chest computed tomography scan reveals consolidation with abscess in lower lobe of the right lung (white arrow) and intrahepatic biloma (black arrow). (B) A tubogram shows a fistulous communication (white arrow head) between the biloma (white arrow) and lower lobe of the right lung (black arrow).

bronchoscopy (FOB) demonstrated the copious bile originated from the right lower bronchus. Preoperative arterial blood gas analysis (ABGA) showed pH 7.41, PaCO₂ 32 mmHg, PaO₂ 67 mmHg and HCO₃⁻ 20.1 mmol/L with room air. The patient's laboratory results revealed leukocytosis (15,300/mm³), total and direct bilirubin of 3.74 mg/dL and 2.30 mg/dL, respectively, alkaline phosphatase of 560 IU/L, γ -glutamyl transferase of 128 U/L, albumin of 2.8 g/dL, lactate of 2.8 mmol/L and international normalized ratio of prothrombin time of 1.57. *Staphylococcus aureus*, *Escherichia coli*, *Enterococcus faecium* and *Pseudomonas aeruginosa* were present in the sputum culture. Surgical treatment was determined because the patient showed gradual deterioration with hypoxemia and uncontrolled pneumonia due to BBF.

On the day of the operation, the patient underwent PTBD for preoperative biliary decompression immediately before being brought to operation room. On arrival in the operating room, the patient was icteric in productive cough with yellowish sputum but did not complain of significant dyspnea in semi-Fowler position. He was continuously monitored by electrocardiogram, pulse oximetry and continuous invasive arterial blood pressure (ABP) in the left radial artery. He revealed oxygen saturation (SpO₂) of 94% in room air, heart rate of 117 beats/min, and ABP of 121/75 mmHg. He was preoxygenated with 100% oxygen via facemask for 5 minutes in a slightly right tilted and reverse Trendelenburg position before the induction of anesthesia. After rapid sequence induction with etomidate 6 mg followed by rocuronium 60 mg, left-sided double-lumen endobronchial tube (DLT, human-broncho, 37 Fr, Wonju, Korea) was inserted. Both the tracheal and bronchial cuffs were immediately inflated.

After correctly positioning DLT by auscultation of breathing sounds and visualization with FOB, both lungs were ventilated with 50% oxygen in air to maintain eucapnia (Table 1). Anesthesia was maintained with target-controlled infusion of propofol and remifentanyl in order to keep bispectral index range of 40 to 60. The patient was monitored for central venous pressure (CVP) with cannulation of right internal jugular vein. Esophageal temperature and urine output were also monitored during surgery.

When trachea and both bronchi were repeatedly suctioned immediately after insertion of DLT, bile-stained secretions were

found primarily in trachea, carina and right bronchus under FOB. Secretions were minimal in left bronchus. Biliary spillage from right lower bronchus was repeatedly suctioned with saline until the fistula tract was ligated (Fig. 3). OLV was initiated with tidal volume of 8 ml/kg, respiratory rate of 15/min, 100% oxygen and positive end-expiratory pressure of 5 cmH₂O in the lateral decubitus position. The patient's SpO₂ was then decreased to 92%, but gradually increased to 97% (pH 7.25, PaCO₂ 47 mmHg, PaO₂ 92 mmHg and HCO₃⁻ 20.6 mmol/L). He underwent repeated bronchial lavage with saline followed by resection of the fistula between the dilated intrahepatic bile duct and basal segment of the right lower lobe. Bilobectomy of the right lung was also performed due to multiple abscess pockets and necrotizing pneumonia in middle and lower lobes (Fig. 3). The perforated diaphragm was debried and repaired primarily around the muscle after insertion of a subphrenic drain.

The patient's oxygenation gradually improved as surgery progressed (Table 1). Eventually two-lung ventilation (TLV) was reinstated after recruit maneuver with 25 cmH₂O for 30 second at the end of surgery. Then, PaO₂ reached 201 mmHg with 60% oxygen. The durations of anesthesia and operation were 445 minutes and 340 minutes, respectively. Despite the estimated blood loss of 1 L, his ABP and CVP remained in the range of 100/60 to 125/70 mmHg and 5 to 10 mmHg, respectively, with administration of 4.2 L of crystalloid, 2 units of packed red blood cells and dopamine at a rate of 3 to 5 μ g/kg/min. Intraoperative urine output was 1480 mL. Hypokalemia due to external biliary drainage in the preoperative period was corrected with intravenous potassium repletion (Table 1). At the end of surgery, the DLT was replaced with a single-lumen endotracheal tube (internal diameter 8.0 mm) for postoperative mechanical ventilation. The patient was transferred to the surgical intensive care unit. He was sedated with intravenous infusion of dexmedetomidine and remifentanyl during controlled mandatory ventilation. Postoperative chest X-ray showed decreased effusion of right lung (Fig. 1). He was successfully extubated on the 4th postoperative day. On follow-up examination for 1 month, there was no sign of BBF recurrence. However, the patient unfortunately succumbed to progressive sepsis, wound infection, hepatic failure and necrotizing pneumonia due to repeated biliary stent stricture 4 months later.

Table 1

Changes in ventilation, oxygenation, circulation, and laboratory tests.

	After anesthesia	pleural incision	After fistulectomy	After Bilobectomy	End of surgery
FiO ₂ (%)	50	100	80	60	60
Ventilation (+ min)	TLV	OLV+25	OLV+110	OLV+230	TLV
PIP (mmHg)	23	27	27	23	24
ABP (mmHg)	112/60	110/59	125/64	109/61	105/58
Heart rate (bpm)	110	105	100	101	110
CVP (mmHg)	8	7	5	6	6
Temperature (°C)	36.8	37.0	37.2	37.3	37.5
pH	7.29	7.25	7.32	7.34	7.34
PaCO ₂ (mmHg)	44	47	41	40	41
PaO ₂ (mmHg)	101	92	160	108	201
HCO ₃ ⁻ (mmol/L)	21.2	20.6	21.1	21.6	21.6
Lactate (mmol/L)	0.6	0.5	0.5	0.5	0.7
Hemoglobin (g/dL)	8.8	9.5	8.5	9.2	9.5
K ⁺ (mmol/L)	2.6	2.8	3.1	3.2	3.3

ABP = arterial blood pressure, CVP = central venous pressure, OLV = one-lung ventilation, PIP = peak inspiratory pressure, TLV = two-lung ventilation.

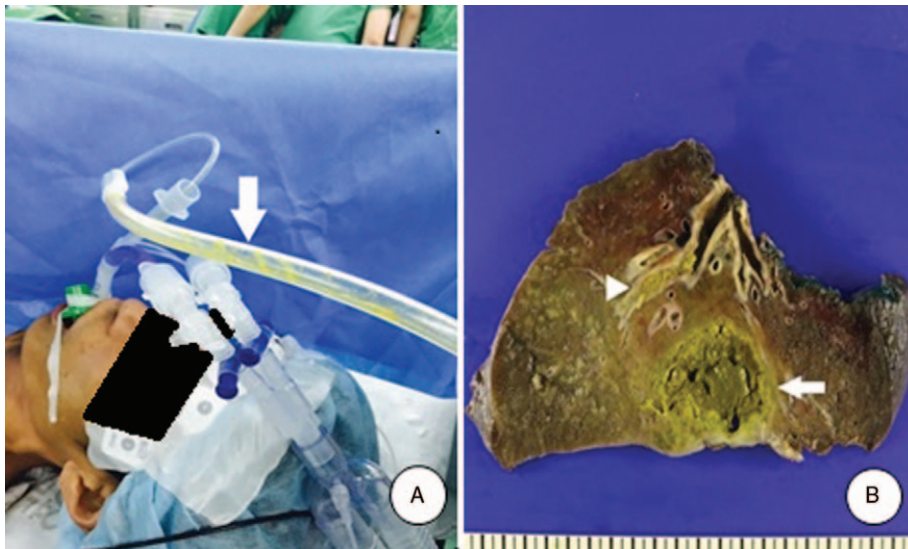


Figure 3. (A) Copious bile-colored secretion (arrow) is suctioned from the right bronchus after left-sided double-lumen endobronchial tube insertion. (B) Lower lobe of the right lung has the infected abscess (arrow) and bronchus filled with bile and infected material (arrow head).

3. Discussion

Since a hydatid cyst of the liver connecting with the lung was first reported by Peacock,^[4] the pathogenesis of acquired BBF may be explained by two mechanisms.^[5] The first is biliary obstruction of different etiologies, which results in retention of bile proximal to the barrier as well as the formation of a biloma and abscess in the liver. A systematic review reported that biliary obstruction is the second most prevalent cause (30.8%) of BBF after hepatic tumors.^[1] The second is a direct invasion of a hydatid cyst or an abscess of echinococcal origin in the liver without biliary obstruction. Irrespective of primary pathology, the cyst or abscess gradually enlarges and directly erodes the diaphragm and/or adjacent lower lobe of the lung until it reaches the nearest bronchus (BBF) or pleural space (pleurobiliary fistula; PBF). Therefore, most fistulae typically involve the basal segments of the right lower lung.^[1,6]

Although biloptysis is pathognomonic for BBF, patients present acute or chronic symptoms such as irritating cough, dyspnea, abdominal pain, jaundice and fever, which resulted from pulmonary pathology combined with underlying hepatic and biliary pathology.^[1] Pneumonia is the most common comorbidity in patient with BBF. Clinical diagnosis is made by the presence of bile in sputum^[7] with suspicious pneumonia. For definite diagnosis, imaging studies including endoscopic retrograde cholangiopancreatography, percutaneous transhepatic cholangiography, magnetic resonance cholangiopancreatography and hepatobiliary iminodiacetic acid scan are necessary to identify the fistula between the bile duct and bronchial tree.^[1,7,8]

The principle of treatment is adequate treatment of intrahepatic and subphrenic abscess, relief of biliary obstruction and correction of underlying cause. Treatment of BBF generally involves medical, surgical and combined approach. Recently, the number of nonsurgical approaches that involve drainage of bile via endoscopic and percutaneous biliary stenting has increased.^[8–10] Surgical approaches with drainage of the subdiaphragmatic abscess with or without resection of the fistula and involved lung should be considered if other medical approaches have failed or when there are complications from underlying

disease.^[3] The combined approach uses drainage of bile (ERBD or PTBD) and abscess (ultrasound or CT- guided) followed by surgical intervention.^[5] Thoracotomy is necessary if preoperative investigation indicates the need for lung resection or in the case of posttraumatic thoracobiliary fistula (BBF and PBF). Laparotomy is mandatory if biliary tract obstruction cannot be managed conservatively.

In our patient, postoperative stricture of the biliary tract after pancreaticoduodenectomy was suspected to have contributed to BBF. The collection of bile resulted in formation of biloma, which subsequently eroded the adjacent diaphragm and adhesive pleura and lung. Because of recurrent biliary obstruction and progressive bronchopneumonia despite conservative treatment, the patient underwent definitive surgical treatment. In this patient, laparotomy for biliary reconstruction was impossible because of anatomic disruption at the hepaticojejunostomy site. Therefore, he underwent preoperative biliary decompression using PTBD followed by thoracotomy including percutaneous drainage of the subdiaphragmatic abscess as well as resection of the fistula and necrotizing lobes of the right lung.

Perioperative management of patients with BBF is a challenge for anesthesiologists because of poor lung function, sepsis and malnutrition before surgery as well as the requirements for lung isolation during surgery and respiratory support after surgery. It is important to preoperatively stabilize the patient's general condition and respiratory function with antibiotic therapy, nutritional support, effective drainage of bile, chest physiotherapy, and incentive spirometry. Fluid and electrolyte imbalance and digestive problems secondary to significant bile loss should also be corrected before surgery.^[1] Acute dyspnea with hypoxemia due to biliary spillage was significantly relieved by preoperative biliary drainage with PTBD in our patient. The major anesthetic considerations for patients with BBF are avoidance of positive-pressure ventilation (PPV) before lung isolation and prompt establishment of the lung isolation of the affected lung to protect the uninvolved lung from contamination as well as ventilation after the induction of anesthesia. Rapid-sequence induction in a reverse Trendelenburg position can avoid further contamination of the healthy lung with

bile and infected material from the affected lung. Airway managements in patients with BBF require meticulous lung isolation, which provides PPV of the healthy lung and prevents flooding of bile from the affected lung to the healthy lung. DLT is generally preferred for lung isolation because it facilitates suctioning of copious bile present in the tracheobronchial tree. Lung isolation in children with BBF can be achieved by using the Fogarty embolectomy catheter.^[11] We confirmed proper placement of the tube by FOB, and determined air tight isolation by monitoring inspired and expired tidal volumes for lung isolation.

Adequate oxygenation in our patient was maintained during surgery despite slight reduction in SpO₂ with institution of OLV. Ventilatory leak should be carefully checked after the completion of surgery. In patients with BBF, respiratory therapy and pain management are the critical components of postoperative care. Continuous thoracic epidural analgesia can provide adequate pain control to ensure good respiratory effort and recovery in the postoperative period.^[12] However, because placement of a thoracic epidural catheter is generally not recommended in patients with sepsis, continuous infusion of intravenous opioid was used for postoperative analgesia in our patient. It is vital to continue chest physiotherapy, bronchodilators, incentive spirometry, postural drainage, and antibiotic treatment in the postoperative period to improve respiratory outcome.^[1]

In summary, BBF is a potentially serious disorder with significant morbidity and mortality. Anesthesiologists should prevent respiratory complications in patients with BBF by improving poor lung function, sepsis and malnutrition before surgery as well as by providing lung isolation for effective oxygenation and bronchial toilet during surgery and by providing respiratory support after surgery.

Author contributions

Conceptualization: Sung Mee Jung.

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Supervision: Sae-Yeon Kim.

Visualization: Yongbae Lee.

Writing – original draft: Jungwon Lee.

Writing – review & editing: Jungwon Lee, Sung Mee Jung.

References

- [1] Liao GQ, Wang H, Zhu GY, et al. Management of acquired bronchobiliary fistula: a systematic literature review of 68 cases published in 30 years. *World J Gastroenterol* 2011;17:3842–9.
- [2] Yoon DH, Shim JH, Lee WJ, et al. Percutaneous management of a bronchobiliary fistula after radiofrequency ablation in a patient with hepatocellular carcinoma. *Korean J Radiol* 2009;10:411–5.
- [3] Jung SI, Goo JM, Han JK, et al. Recurrent bronchobiliary fistula: unsuccessful management with repeated insertion of metallic biliary stent. *J Vasc Interv Radiol* 2003;14:1577–9.
- [4] Peacock T. Case in which hydatids were expectorated and one of supuration of a hydatid cyst of the liver communicating with the lungs. *Edinburgh Med and Surg J* 1850;74:33–46.
- [5] Crnjac A, Pivec V, Ivanec A. Thoracobiliary fistulas: literature review and a case report of fistula closure with omentum majus. *Radiol Oncol* 2013;47:77–85.
- [6] Boyd DP. Bronchobiliary and bronchopleural fistulas. *Ann Thorac Surg* 1977;24:481–7.
- [7] Gugenheim J, Ciardullo M, Traynor O, et al. Bronchobiliary fistulas in adults. *Ann Surg* 1988;207:90–4.
- [8] Rose DM, Rose AT, Chapman WC, et al. Management of bronchobiliary fistula as a late complication of hepatic resection. *Am Surg* 1998;64:873–6.
- [9] Pinsker N, Papoulas M, Sodergren M, et al. Successful endoscopic management of a persistent bronchobiliary fistula with Histoacryl[®]/Lipiodol[®] mixture. *Ann R Coll Surg Engl* 2018;100:e73–7.
- [10] Shim JR, Han SS, Park HM, et al. Two cases of bronchobiliary fistula: case report. *Ann Hepatobiliary Pancreat Surg* 2018;22:169–72.
- [11] Chandran R, Batra RK, Agarwala S, et al. Selective bronchial blockade with Fogarty catheter in a child with acquired bronchobiliary fistula. *Paediatr Anaesth* 2013;23:373–5.
- [12] Mitra S, Bhatia N, Dey N, et al. Bronchobiliary fistula: an anesthetic challenge!. *J Clin Anesth* 2009;21:360–2.