A fistulous communication between the bile ducts and bronchial tree (bronchobiliary fistula) is a rare condition. It can be congenital or can occur secondary to thoracoabdominal trauma, liver abscesses, hepatic hydatid disease, choledocholithiasis, surgery or hepatobiliary neoplasms. Bronchobiliary fistula poses a difficult and complex management problem and surgery has been considered as the most appropriate therapeutic option. Recent innovations in minimally invasive interventions have led to successful use of percutaneous or endoscopic approaches in patients of bronchobiliary fistulas; however, the experience is limited to few cases only. We present a rare case of combined bronchobiliary and bilio-cutaneous fistula which developed following right hepatectomy for blunt trauma to the abdomen. The patient was successfully treated with prolonged biliary drainage.

Case Report

A 22-year-old man underwent right hepatectomy at a tertiary care hospital for extensive laceration of the liver after being hit by a cricket ball in the upper abdomen. The patient had an uneventful postoperative course. However, even after 4 weeks of surgery, he continued to drain more than 500 mL/day of bilious fluid from the sub-hepatic drain placed during surgery. Subsequently, the patient was referred to us for endoscopic retrograde cholangiopancreatography (ERCP). The patient came to us 6 weeks after surgery with a daily drain output of 500–800 mL of clear bilious fluid. There were no other symptoms and clinical examination was normal. Hematological and biochemical investigations were within normal limits. Contrast-enhanced computerized tomography (CECT) of the abdomen did not reveal any intra-abdominal collection.

An informed consent was obtained and ERCP was done using duodenoscope (TJF-160, Olympus Corp., Tokyo). The cholangiogram revealed normal-sized bile duct and biliary radicals in the left lobe of liver. There was extravasation of contrast from multiple sites in the bile duct suggestive of bile leak from multiple sites (Figure 1). Over a 0.035-inch Jag guide wire (Microvasive, Boston Scientific, Mass) a 7-Fr Amsterdam type biliary stent was placed in the bile duct below the disruption, as repeated attempts to place the guide-wire across the disruption failed and the guide-wire went into the disruption repeat-
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edly. Following this, the patient had a gradual clinical improvement and the drain output subsided after 4 weeks of ERCP. At this stage an ultrasound of the abdomen did not reveal any intra-abdominal collection. The abdominal drain was removed.

After an asymptomatic period of 2 weeks, the patient returned to us with complaints of bilious discharge from the site of abdominal drain and cough with bile colored sputum (bilioptysis). He did not have any fever or abdominal pain. A small subphrenic collection was seen on abdominal ultrasound. On fluoroscopic examination of the abdomen, the biliary stent could not be seen, suggestive of spontaneous migration of the stent. The patient was admitted and started on intravenous cefotaxime and metronidazole. Pressure injection of the contrast from the cutaneous site of the bile leak (fistulogram) demonstrated the pooling of contrast in the subphrenic space, with contrast tracking upwards demonstrating communication with the right bronchial tree (Figure 2). The delayed fluoroscopic pictures demonstrated the right main bronchus delineated by the contrast (Figure 2). The patient was taken up for ERCP and a 1 cm biliary sphincterotomy was performed and a 7-Fr nasobiliary drain (NBD) was placed below the disruption. Following this, the patient had dramatic improvement and both the bilioptysis and cutaneous discharge subsided. However, after 10 days of ERCP the NBD got accidentally pulled out and the patient had recurrence of bilioptysis and cutaneous discharge. Because of a complex bile ductal injury, it was decided to place multiple transpapillary biliary stents. The patient was taken up for repeat ERCP and cholangiogram revealed persistence of bile leak, but the amount of leak had markedly reduced with a single site of leak seen in contrast to multiple sites of leak in the earlier cholangiogram (Figures 2 and 1). Two 10 Fr stents were placed below the disruption. Following this, both the bilioptysis and external biliary drainage subsided and no intra-abdominal collection was seen on ultrasound done 4 weeks after ERCP. After 5 months of follow-up, there has been no recurrence of biliocutaneous fistula or bilioptysis.

Discussion

Bronchobiliary fistulas have been traditionally managed surgically by either using a combined thoracoabdominal approach for repair of the fistula and repair of ductal injury or an abdominal approach to relieve the bile duct obstruction.4 With availability of safe and effective minimally invasive interventional techniques, bronchobiliary fistulas have also been successfully treated with percutaneous, bronchial and endoscopic methods.1,2,3,5

Endoscopic transpapillary biliary drainage in the form of biliary sphincterotomy and / or biliary stenting or nasobiliary drainage has been shown to be highly effective in the treatment of postoperative and post-traumatic biliary leaks.6,7 Following these encouraging results, endoscopic therapy has also been tried in the management of bronchobiliary fistulas with good results; however, published experience is limited and there is no consensus on the type and duration of therapy.2,3,8,9 There are published reports about successful treatment of bronchobiliary fistulas with nasobiliary drainage or endoscopic stenting with or without sphincterotomy and majority of these fistulas have been reported to be secondary to hydatid disease of liver.2,3,8,9 Because of complex nature of these fistulae, healing takes a longer time and recurrence of fistula after successful endoscopic therapy has been reported.3 As a result, prolonged drainage may be needed for successful outcome. Katsinelos et al10 highlighted the role of prolonged endoscopic drainage and reported a patient who had recurrence of bronchobiliary fistula 6

Figure 2: Fistulogram reveals pooling of the contrast in the sub-hepatic space with subsequent tracking upwards with communication with the right bronchial tree (arrow) (Left panel). Delayed picture of fistulogram reveals delineation of the right main bronchus (Middle panel); Repeat ERCP reveals decrease in the amount of bile leak and a single site of leak is seen (arrow) (Right panel)
months after initial successful surgical repair and resection of right anterior lower lobe. The patient was successfully treated with endoscopic sphincterotomy and insertion of a 11.5 Fr transpapillary biliary stent. However, the fistula recurred after the stent was removed; thereafter the patient was successfully treated with repeated stent exchanges every 6 months for a total period of 30 months.

In conclusion, we have described a rare case of post-traumatic combined bronchobiliary and biliocutaneous fistula successfully treated with endoscopic sphincterotomy and prolonged biliary drainage.

References


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Image

Amyand hernia

A 67-year-old man presented with a right supra-inguinal bulge and pain for the past one day. Physical examination indicated strangulated inguinal hernia. Surgical exploration revealed acute appendicitis within the sac of indirect inguinal hernia (Figure). Appendectomy and Bassini-type herniorrhaphy were performed successfully.

The presence of the appendix within an inguinal hernia has been referred to as “Amyand hernia” to honor Claudius Amyand, surgeon to King George II, who first described it in a 11-year-old boy, and performed a successful transherniotomy appendectomy in 1735.1

This entity usually presents with features of an obstructed or strangulated inguinal hernia. Acute appendicitis or perforation of appendix within the sac simulates intestinal perforation within the hernia. Since it does not have specific symptoms or signs, it is difficult to diagnose Amyand hernia preoperatively.1 The occurrence of herniated appendices is mostly reported in a right inguinal hernia, probably as a consequence of the normal anatomical position of the appendix and also because right-sided inguinal hernias are more common than left-sided hernias.1,2 The presence of appendiceal inflammation or perforation is a contraindication to mesh placement.1

References


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Figure: Acute appendicitis located in the sac of indirect inguinal hernia