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Broncho-Biliary Fistula Due to Hydatid Disease: Case Report

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Abstract

A Broncho Biliary Fistula (BBF) is a rare complication of echinococcosis. This occurs from rupture of a hydatid cyst located at the upper surface of the liver into the bronchial tree. Fistulisation, results in biliopytsis, which is expectoration of bile-tinged greenish-yellow sputum, a pathognomonic feature of BBF. Bilopytsis may also occur in other conditions such as hepatectomies, liver abscesses or trauma. Surgical fistula closure is the definitive management of these patients, however downstream biliary obstruction should be ruled out first. Almost all cases can be approached through a right postero-lateral thoracotomy followed by phrenotomy. However, if there are multiple cysts in the liver, a thoraco-abdominal incision may be necessary.

Keywords: Bronchobiliary fistula (BBF); Bilioptysis; Hydatid disease

Introduction

Human echinococcosis is a zoonotic infection caused by the genus Echinococcus. Primary hosts are the members of the Canidae family, usually dogs, wolves, and coyotes. They contract echinococcosis by ingesting mature, productive echinococcal cysts in the viscera of an intermediate host (e.g., sheep, goats, cattle, hogs, moose, reindeer, deer, elk, etc.). Humans act as an accidental intermediary host who acquire the disease by ingestion of food or water that contains the eggs of the parasite or by close contact with an infected animal [1]. Hydatid disease and complications related to hydatid cysts are therefore common in areas where herding is part of a daily life which is the case in most parts of Africa. A rare complication of this disease is a bronchobiliary fistula (BBF) a communication between the biliary tree and the bronchial tree. The cardinal manifestation of BBF is the presence of bile in the sputum, known as bilioptysis. While BBF secondary to Liver Hydatid cyst rupture is the most common cause of bilopytsis, approximately 20% cases occur due to other sub diaphragmatic pathologies (Table 1). Here we present a case of BBF secondary to a ruptured liver hydatid cyst successfully repaired through a right posterolateral thoracotomy.

Case Report

A 32-year-old male farmer presented with progressive right upper quadrant abdominal swelling. Six weeks prior to presentation he developed a sudden onset of cough productive of copious amounts of watery and salty sputum. This was followed by expectoration of moderate amount of greenish yellow sputum. Subsequently, the swelling in his abdomen regressed. There was no history suggesting biliary obstruction such as yellowish discoloration of the eyes, cola coloured urine, pale stool or pruritis. No prior surgery or significant medical history. On physical examination, he appeared chronically unwell, with slight tachycardia but no objective fever. Chest Auscultation revealed crepitations on the right anterior lower lung field but no other additional abnormal breath sounds. Abdomen was flat with no sign of organomegaly or ascites. The patient's laboratory investigations were within normal limits apart from a mild degree of anaemia. Abdominal ultrasound was performed, showing no evidence of obstruction to the biliary tree. However, there was a ruptured Hydatid cyst identified in the right lobe of the liver. Chest X-ray showed bronchiectatic changes in the area of right middle lobe. CT scan of the chest and abdomen revealed a ruptured hepatic hydatid cyst communicating with the middle lobe bronchus of the right lung. There was some haziness in the right lower lobe, possibly aspiration of biliary contents (Figures 1-4). The fistula

Causes of Bronchobiliary fistula
Trauma
Lithiasis in biliary trees
Subdiaphragmatic abscess
Cholecystitis/pancreatitis
Liver/Biliary tree tumor
Radiofrequency ablation
Hepatic resections
Necrotizing lung infections

Table 1: Non-hydatid cyst causes of Bronchobiliary fistula.

Figure 1: CT of the chest showing consolidated and bronchiectatic segment of the right middle lobe.

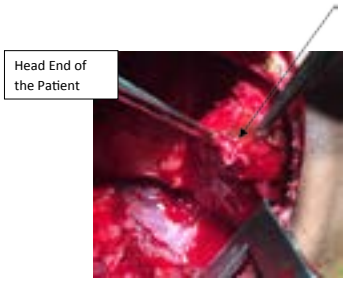

Figure 2: The fistulous tract divided through right posterolateral thoracotomy, View of diaphrgam with biliary fistula coming through (Arrow).

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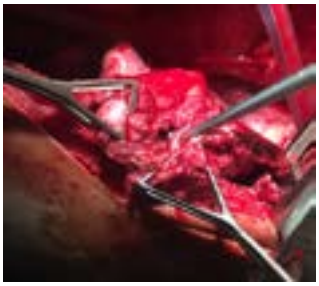

Figure 3: The fistulous tract on the lateral segment of the Right middle lobe of the lung.

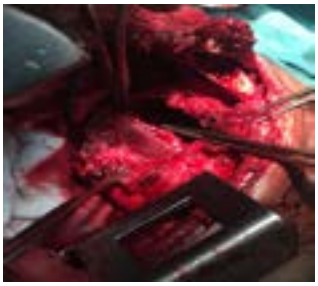

Figure 4: Radially opened diaphragm [narrow arrow] showing the cavity in the liver (wide arrow) after excising part of the pericyst.

was divided, and a middle Lobectomy was performed since the lobe was bronchiectatic and not salvageable. The middle lobe bronchus closed in two layers with 5-0 prolene sutures. The fistula tract on the diaphragm was resected and then the diaphragm was opened radially to visualise the liver. The pericyst of the Hydatid cavity was dissected off the diaphragm on the peritoneal side. The cavity was laid open; and the contents were evacuated, including the endocyst. The pericyst was partially excised. There were no identifiable biliary holes nor was there was any apparent biliary leak. Subsequently, the edges of the pericyst were marsupialized. Before closure of the diaphragm a drain was left in the sub-diaphragmatic space and brought out through the right subcostal region. The thoracotomy was closed over a chest drain and the patient was transferred to a high dependency unit for 24 hours. Antibiotic was continued for 7 days post op. The next day he was transferred to the regular ward with stable vital signs. The chest drain was removed on the 6th post-operative day when the lung was fully expanded, and the drainage was serous and <50 ml/24 hrs. The patient was kept in the ward for an additional 26 days due to the subcostal drain was draining significant amounts of bile tinged fluid. Over time, the output progressively decreased and stopped. Subsequently, the patient was discharged. During his 8 months post op follow up he was symptom free and chest X-rays show no residual lung lesion or complication.

Operative Findings and Treatment

The patient was then started on IV Ceftriaxone 1 gm Bid in preparation for a lower right posterolateral thoracotomy. After mobilizing the lung with slight difficulty, the fistula identified between the lateral segment of the middle lobe and the dome of the liver through the muscular diaphragm.

Discussion

Bronchobiliary fistula (BBF) is defined as an abnormal communication between the biliary system and bronchial tree. In 1850, Peacock described the first case of BBF in a patient presenting with liver hydatidosis. BBF has also been reported as a congenital malformation; however, in most cases, it occurs following liver pathology, particularly parasitic infections like echinococcosis [1]. Two major mechanisms are suggested for the development of BBF. One mechanism involves a subphrenic or intrahepatic abscess with inflammatory reaction in the sub diaphragmatic space and subsequent rupture into the bronchial system. The second mechanism describes a mechanical liver pathology (with or without bile duct obstruction) which erodes the diaphragm leading to a communication between bronchial tree and billiary channels [2-4]. Hydatid disease, categorized in the second group, usually is not associated with billiary obstruction in these patients. The fistula in these cases occurs due to a combination of pressure erosion from an expanding Hydatid cyst to the diaphragm and the destructive effect of superimposed infection. If enough adhesions precede the erosion, the cyst will rupture into the pulmonary parenchyma causing pneumonitis and BBF [5,6]. A fistula between right hemi diaphragm and nuda hepatis is the most frequently reported location, while a Bronchobiliary fistula to the left lung has been reported in a Systematic review of 68 cases published in 30 years [7]. While the commonly involved part of the lung is the right lower lobe, the middle lobe can also be involved.

Bilioptysis (bile stained sputum) (ranging in quantity between 100 and 450 ml/ day and presence of bile in a pleural effusion, are both pathognomonic of the existence of a Bronchobiliary fistula. Once BBF develops most frequent symptoms include fever, irritating productive cough, chest pain, right upper abdominal pain and jaundice [2,5,7]. Bile behaves as a strong irritant when present outside the billiary channel and gastrointestinal system. Early diagnosis requires a high index of suspicion, and can be confirmed by sputum analysis. Early detection is important to prevent irreversible lung destruction. Biliary irritation accounts for the majority of the mortality and morbidity from Hydatid BBF, Mortality ranging from 9.6%-2.2% [4,5,8]. This may present as pneumonia or can also present with severe sepsis or even ARDS.

Another form of communication between abdomen and chest, the so called pleurobiliary fistula causes dry and irritating cough, chest pain and findings from the right chest, as result of bile accumulation in the right pleura and development of right basilar atelectasis. Pleurobiliary fistula is more difficult to diagnose unless even a higher degree of suspicion exists, in a patient with predisposing factors. Bronchoscopy has a limited role in the diagnosis and management of a BBF since it is often difficult to visualize the fistula due to the brisk flow of bile and the small size of the tract coupled with the fact that the commonly involved part of the lung is the right lower lobe. Presently, bronchoscopy is mainly used for bronchial toilet [8]. Chest X-ray may demonstrate lower lung zone densities, paracardiac infiltrates, pleural effusions or an atelectasis of the lower pulmonary lobe associated with an elevation of the right hemi diaphragm. Air-fluid level suggesting abscess formation may also be seen but it fails to demonstrate the fistula [8]. Ultra-sonography is an important tool in diagnosing the sub diaphragmatic pathology and ruling out billiary obstruction. However, both modalities fall short in demonstrating the fistula. Thoracic and upper abdominal CT appears to be the investigation of choice in the initial evaluation of a suspected BBF. Although it rarely depicts the BBF, it may provide indirect findings such as subphrenic fluid collection, discontinuity of the diaphragm, bronchiectasis, atelectasis, or a pleural effusion [5,6].

Once BBF is confirmed, it is very important to rule out biliary obstruction, as a BBF will not heal unless this obstruction is relieved. Contrast-enhanced magnetic resonance cholangiography (MRC) is often able to clearly delineate the leakage of contrast agent from the billiary duct and its communication with the bronchial tree. MRC also provides functional information about billiary flow as well as display of

billiary anatomy [9,10] D'Amora Marilina et al. advocated integrated imaging using Contrast-Enhanced Computed Tomography (CECT) and Magnetic Resonance Cholangiography (MRC) for definitive diagnosis of a BBF [11,12]. Hepatobiliary scintigraphy, which is used to visualize the liver and biliary tree, has also been used as a non-invasive means for the precise diagnosis of a BBF [13].

Endoscopic retrograde cholangiopancreatography (ERCP) and Percutaneous transhepatic cholangiography (PTC) may be performed for evaluation of the biliary anatomy and BBF. ERCP also offers a therapeutic option in those circumstances where there is a biliary obstruction [4]. However, it is not recommended as a purely diagnostic test, due to the invasive nature of the investigation and the reliability of alternative imaging techniques such as MRC. [5,9,10].

The goal of the management of BBF is correction of the possible biliary obstruction with adequate drainage of any concomitant abscess and treatment of the underlying disease. Despite the increasing number of reported cases, though, a widely accepted strategy for the management of BBF remains to be defined [4]. The traditional management of BBF is surgery. However less invasive modalities have been described and are getting ground. Endoscopic treatment of BBF has been reported only as a first stage treatment until the patient's condition stabilizes. Kima et al. had reported histoacryl embolization under Bronchoscopy guidance, for a 56-year-old patient who had a recalcitrant BBF following treatment for hepatocellular cancer. This patient had failed arterial chemotherapies, radiofrequency ablations, chemoembolization, and a right lobectomy. The bilopytsis has resolved after histoacryl embolization, but patient died from biliary obstruction and hepatic failure three months later [7,14].

Endoscopic placements of biliary stents, as well as endoscopic sphincterotomy have been described for BBF, in an attempt to relieve the downstream obstruction. However, these less invasive treatment modalities usually involve prolonged hospitalization and multiple image-guided procedures. Despite multiple attempts at non-invasive therapies, the patient may ultimately return to the operating room for more definitive treatment [15]. Surgery still remains the treatment of choice in cases of BBF secondary to Echinococcosis. The approach, most of the time, is right postero-lateral thoracotomy, however in few cases a thoraco-abdominal approach may be mandatory to access complex and multiple cysts [6,16]. A two-stage approach can be used as suggested by Gugenheim [2]. This involves (1) external biliary drainage by percutaneous means or by direct surgical drainage of the overlying sub phrenic abscess and biliary tract and (2) treatment of the underlying cause. In patients with biliary obstruction, the priority is treatment of the biliary disease. This is best achieved by a transabdominal approach [16]. The principles according to Ferguson and Burford [17] for successful management of a BBF include:

- Early aggressive treatment by thoracotomy.
- Adequate subcostal drainage of the hepatic bed under direct vision.
- Secure closure of the diaphragmatic perforation by non-absorbable sutures.
- Decortication for the lung.
- Lobectomy for Broncho biliary fistula.

Extended pulmonary resections are generally not recommended given the significant risk of recurrence of echinococcosis. Therefore, parenchymal sparing operations are usually preferable. However, in situations where there the underlying lung is not salvageable, an anatomic segmentectomy or lobectomy may be necessary, which was the case in our patient where the right middle lobe was bronchiectatic and had to be resected [3,6,8,12,18].

Conclusion

Our patient presented with a case of ruptured liver Hydatid cyst creating BBF. CT scan demonstrated bronchiectatic changes in the right middle lobe although the fistula could not be directly demonstrated. He had no clinical or radiologic signs of biliary obstruction, which is commonly seen in BBF secondary to ruptured Hydatid cyst. The BBF was managed by right middle lobectomy, resection of the tract, and definitive management of the hydatid cyst in a transdiaphragmic manner.

BBF secondary to ruptured hydatid cyst of the liver is a rare event. Surgery arguably is the best management modality.

References

1. Thomas W, LoCicero J, Reed C, Richard H, Feins E (2009) General Thoracic Surgery (7th edn) Shields Lippincott Williams & Wilkins, USA.
2. Gugenheim J, Ciardullo M, Traynor O, Bismuth H (1988) Bronchobiliary fistulas in adults. Ann Surg 207: 90-94.
3. Heidi KC, Allen MS, Deschamps C, Miller DL, Pairolero PC, et al (2000) Bronchobiliary fistula: Principles of management. Ann Thorac Surg 70: 1392-1394.
4. Esme H, Fidan F, Orman A, Unlu M, Acar M, et al. (2005) A case of bronchobiliary fistula associated with subdiaphragmatic abscess after hepatic surgery. IJTCVS 21: 177-179.
5. Nikolaos K, Nikolaos P, David L (2014) The challenging management of hepatopulmonary fistulas. J Thorac Dis 6: 1336-1339.
6. Eryigit H, Oztas S, Urek S, Olgac G, Kurutepe M, et al. (2007) Management of acquired bronchobiliary fistula: 3 case reports and a literature review. J Cardiothoracic Surg 2: 52.
7. Liao GQ, Wang H, Zhu GY, Zhu KB (2011) Management of acquired bronchobiliary fistula: A systematic literature review of 68 cases published in 30 years. World J Gastroenterol 17: 3842-3849.
8. Gerazounis M (2002) Bronchobiliary fistulae due to echinococcosis. Euro J Cardio-Thoracic Surg 22: 306-308.
9. Karabulut N, Çakmak V, Kiter G (2010) Confident diagnosis of bronchobiliary fistula using contrast-enhanced magnetic resonance cholangiography. Korean J Radiol 11: 493.
10. Fulcher AS, Turner MA, Capps GW (1999) MR cholangiography: Technical advances and clinical applications. Radiographics 19: 25-44.
11. Marilina D (2015) Integrated Imaging in a bronchobiliary fistula after radiofrequency ablation treatment J Clini Case Rep 1: 012.
12. Kumar P, Kabra S, Mehta P, Ismail J, Agarwala S, et al. (2015) Brocho-biliary fistula: A rare complication after ruptured liver abscess in a 3½ year old child. Lung India 32: 489.
13. Mohammadreza S, Kiani M, Javadi H, Raeisi A, Assadi M (2015) Hepatobiliary scintigraphy with SPET in the diagnosis of bronchobiliary fistula due to a hydatid cyst. Hell J Nucl Med 18: 160-162.
14. Jung HK, Kim MD, Lee YK, Hwang SG (2008) Bronchobiliary fistula treated with histoacryl embolization under bronchoscopic guidance: A case report. Respiratory Medicine CME 1: 164-168.
15. Moumen M, El Fares F (1991) Bilio-bronchial fistula of hydatid origin. Apropos of 8 Cases. J Chir (Paris) 128: 188-192.
16. Tocchi A, Mazzoni G, Miccini M (2007) Treatment of hydatid bronchobiliary fistulas: 30 years of experience. Liver Int 27: 209-214.
17. Ferguson TB, Burford TH (1967) Pleurobiliary and bronchobiliary fistulas. Surgical management. Arch Surg 95: 380-386.