



Biliothorax: Review of 3 Centers Experience

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Abstract

Introduction: Biliothorax is the accumulation of bile in the pleural cavity. Congenital bronchobiliary or pleurobiliary fistulas, hydatid disease and or liver abscess whether echinococcosis and or amoebic abscess, pyogenic, bile tracts obstruction, blunt and or penetrating thoraco-abdominal trauma, liver resection, radiofrequency ablation, bile duct stricture and radiotherapy may be the accused aetiology of such entity. This study aims at reviewing our experience in King Abdullah Medical City; Makkah, King Fahad Central Hospital Jazan; Saudi Arabia and Arar Central Hospital; Saudi Arabia in the management of cases of biliothorax in the last 7 years.

Patients and Methods: The files of 36 cases of biliothorax were reviewed including the available data: age, sex, main presentation, diagnosis, investigations, different modalities of treatment, results and follows up if any.

Results: Thirty six patients were included in this retrospective study. Females were 20, age varied between 16 and 82 years. The aetiology was hepatobiliary tumors in 8, sub-phrenic abscess in 7, and trauma in 7 cases; four of whom were iatrogenic and liver abscess in 6. The main presentation was persistent greenish chest tube drainage of a large amount. The main investigation used was chest x ray, abdominal US, ERCP and MRCP. Interventional radiologist was consulted in 18 cases. Abdominal surgery was required in 11 cases. Thoracic surgery was performed in 7 cases. Four cases died during the study interval.

Conclusion: Biliothorax is a true dilemma mandating a multidisciplinary approach in diagnosis and management. It carries a high morbidity, recurrence, failure of treatment and mortality.

Keywords: Biliothorax; Truma; Drainage

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Introduction

Many designations may point to the same problem such as bronchobiliary, pleurobiliary, biliobronchial, biliary-bronchial, bilio pulmonary, hepatobronchial, cholecystobronchial, broncho-hepatico-cystic and broncho-pleuro-colonic fistula and pleurobilia, biliothorax, tracheocholedochal tract, bilitracheal fistula with trifurcation of the bronchi and carinal trifurcation with tracheobiliary fistula. For the purpose of simplification, thoracobiloma is both descriptive and simple [1-13]. Biliothorax is also but rarely called cholethorax. Two mechanisms may be blamed as a pathogenesis of biliothorax. Liver biloma and abscess with or without obstruction. Bile will erode the diaphragm especially at the posteromedial part of the right hemi diaphragm which is in a direct contact with the area porta hepatis [1-3]. It is a serious condition with a high mortality rate and requires a well-planned multidisciplinary management strategy [1,7]. Biliothorax can be caused by hepatic hydatidosis, amebiasis, liver abscesses, gallbladder disease with or without cholelithiasis, iatrogenic perforation during PTC, PTBD, open cholecystectomy, liver biopsy, and bilioenteric bypass, blunt hepatic trauma, penetrating thoracoabdominal trauma, and rarely the cause is still unknown. It occurs when the barriers between the hepatobiliary system and pleural space are broken. Interestingly, cholethorax was reported to occur with or without biliary-pleural fistula [8-13]. Biliothorax is suspected when there is a large amount of greenish drainage from the pleura. Diagnosis is confirmed by pleural fluid/serum bilirubin ratio more than 1. It has been suggested that if thoracentesis is delayed, the pleural fluid biochemistry may be non diagnostic. This may explain the failure to see an elevated pleural/serum bilirubin ratio on the second thoracentesis. Only with recurrent biliary leakage were the laboratory results confirmatory of a bilious effusion [11-13].

Patients and Methods

The files of our patients with biliothorax were reviewed thoroughly regarding the history, general, local examinations laboratory results and radiological investigations needed. The management lines were also reviewed. The OPD clinic visits were reviewed. Biliothorax was suspected if the drainage was excessive and was greenish in color. It was confirmed if the pleural fluid bilirubin/ serum bilirubin ratio was more than 1. Our rationale of treatment is similar to the standard management of any fistula, in that all fistulas will eventually close if well treated any associated infection, malignancy or distal obstruction. Chest tube or pigtail was inserted. Fluid was sent for analysis, culture sensitivity, chemistry, bilirubin level, although the optimal treatment of PBF is still debated. Although a minimally invasive approach is preferred because of the high morbidity associated with thoraco-abdominal surgery, we never attempted it in our cases. Chest tube was inserted and sometimes pigtail was inserted in the pleural space. US examination of the pleural cavity, liver, hepatobiliary tract, subphrenic spaces was routinely performed. CT chest and abdomen with contrast and MRCP, ERCP were usually performed to delineate the pleural cavity, diaphragm, subphrenic spaces, hepatobiliary tract and other pathologies. The management varied between just drainage of the pleural space under local anesthesia and complete antiseptic techniques. Drainage of the subphrenic abscesses was performed either by an open or transcutaneous techniques. ERCP sphincterotomy with or without stenting was done by the endoscopist if there is a stenosis in the biliary tract with post stenotic dilatation. Laparotomy was performed to excise hydatid cysts, resect tumors or liver lobes, put a T tube and or bilio-enteric anastomosis was the last resort in cases of failed or refractory hepatobiliary obstruction with persistent biliothorax despite all the conservative and or interventional tools. Thoracotomy was required for decortication, lobe resection and hydatid cyst resection. Follow up of patient was through the outpatient clinic with follow up of the symptoms, radiology and management of any complaints.

Results

Our retrospective study included 36 cases of biliothorax. Twenty females were included (55.55%) (Table 1). Age varied between 16 and 82 years with the median age of 41 years. The main presentation was greenish effusion, bilioptysis, fever, jaundice, pain and fever. Bilio pleurocutaneous fistula was the rarest presentation in our patients; only 1 patient (Table 2). The chest tube duration varied between 20 days to 120 days with a median duration of 43 days. The aetiology included hepatobiliary malignancy, trauma, liver abscess, biliary stenosis, echinococcosis, and rarely chronic pancreatitis (Table 3). Chest tube and or pigtail were inserted in all patients as diagnostic and therapeutic. Greenish effusion of a large amount was the main tool to diagnose biliothorax. Conservative treatment was needed in 28 cases. It involved drainage of the pleural cavity + ERCP intervention with or without percutaneous drainage of the liver or sub-phrenic abscesses. ERCP was used to dilate the stenotic or obstructed hepatobiliary tracts with or without stents (Table 4). Open surgery was needed in 18 cases; 11 laparotomies and 7 thoracotomies. Two patients needed laparotomy followed by thoracotomy few weeks later. Fourteen patients needed more than one line of treatment. We started with chest tube which was successful in only 4 cases. Then according to the CT, MRCP and ERCP, the line of further management was followed. Five cases needed chest tube insertion followed by laparotomy followed by thoracotomy. During our study, 4 patients died, three from advanced

Table 1: Sex distribution in our cohort.

Sex	No.
Females	20
Males	16

Table 2: Presentation.

Main Symptom or sign	Number
Greenish effusion	32
Expectoration of either frank bile , Questionable bile content, Hydatid debris, Amebas or Purulent mucus	28
Fever	24
Jaundice	24
Tender hepatomegaly: right shoulder, right lower chest pain or right upper abdominal pain	15
Dyspnea	14
Bilioleurocutaneous Fistula	1

Table 3: Aetiology

Hepatobiliary Malignancy	8
Subphrenic Abscess	7
Trauma	7
Iatrogenic	4
Non Iatrogenic Trauma	3
Liver Abscess	6
Biliary Stenosis	4
Hydatid	3
Chronic Calcific Pancreatitis	1

Table 4: Management.

Chest Tube	32(88.88%)
Pig Tail	4(11.11%)
ERCP	18(50%)
Percutaneous Drainage	10(27.77%)
Liver Abscess	5(13.88%)
Subphrenic Abscess	5(13.88%)
Laparotomy	11
Hydatid Cyst	3
Tumor Resection	3
Liver Lobectomy	2
T Tube	2
Bilioenteric Anastomosis	1
Thoracotomy	7
Decortication	4
Lobectomy	2
Hydatid Cyst Resection	1

malignancy and one from severe malnutrition and debilitation. One patient developed pleuro-bilio-cutaneous fistula and one developed entero-bilio-cutaneous fistula.

Discussion

We are presenting 36 cases of biliothorax which is a good number for such a problem. Gugenheim et al. [5], reported 16 cases diagnosed as biliothorax due to fistula in adults observed over 32 years. Singh et al. [14], reported 8 cases in 5 years while Eryigit et al. [6], reported 3 cases in 2007. The principal aetiology was trauma whether blunt and or sharp. Conservative treatment or negligence of hepatobiliary trauma is a major co-factor [3,15]. Females dominated our series. In the series Liao et al. [16], the median age of on presentation was 48.3 years (range, 14-87 yrs). Our series included patients whose age varied between 16 and 82 years with the median age of 41 years. Hepatobiliary tumors followed by Trauma whether iatrogenic or non iatrogenic followed by abscesses whether subphrenic or liver, followed by biliary

stenosis were the main pathologies in our series. The rarest causes in our study were hydatid and chronic pancreatitis. Hepatobiliary tumors in our series were cholangiocarcinoma, hepatocellular carcinoma and metastatic liver tumors. Over 2/3 of our cases were right sided biliothorax. This copes with Ragozzino et al. [17], findings. Most of the cases of Liao et al. [16] were right sided. A high index of suspicion is mandatory for early diagnosis which is crucial for the management of PBF. Delayed management will be very hazardous as bile irritates the lung, pleura and skin and if it persists, it can cause a major lung injury and damage including fibrosis, pneumonitis and or lung destruction. Diagnosis is made primarily on clinical history symptoms and signs [18,19]. In our series, a large amount of dark greenish pleural drainage alerts us to ray the red flags to the diagnosis of biliothorax. Biliothorax was confirmed by pleural/serum bilirubin ratio more than 1. This was agreeing with the practice of other groups such as Al-Qahtani [7] and others [11-13]. Green pleural effusion of a large amount was the main presentation. It was reported in 32 cases. Four patients presented with a small amount of pleural effusion with other symptoms. Expectoration of bile (biliptysis), hydatid debris, amoebas and purulent mucus in 28 cases, fever in 24, jaundice in 24, enlarged tender right upper quadrant in 15, dyspnea in 14 and bilio-cutaneous fistula in 1. This is similar to those reported by Singh et al. [14] MRCP remains the investigation of choice to identify the cause and site of any biliary obstruction and delineate the tract. ERCP and Percutaneous Transhepatic Cholangiography (PTC) are useful for diagnostic and therapeutic purposes in cases of PBF due to cholelithiasis. ERCP enables us to insert stents, dilate the biliary tracts, remove stones and do sphincterotomy [17-20]. Chest x-ray in 36 cases, abdominal US in 34, MRCP in 27 cases and ERCP in 18 cases. Artunduaga et al. [21], reported cholescintigraphy with SPECT as a valuable diagnostic tool when biliothorax is suspected, because it can help both diagnosing and localizing the fistulous tract. The use of SPECT may obviate the need for delayed planar imaging performed in some cases, and thereby provide a more rapid diagnosis of bronchobiliary fistula [22]. We do not have this facility in our centre. We needed thoracotomy in 7 cases. Pleural decortication and pleurodesis was performed in 2 cases or persistent refractory collapse, collection and extensive adhesions, bi-lobectomy in one patient, right lower lobectomy in one patient and hydatid cyst resection was required in one patient. The standard treatment of any fistula was followed strictly. Although the optimal treatment of PBF is still debated, a minimally invasive approach is preferred because of the high morbidity associated with thoraco-abdominal surgery. No guidelines exist for the treatment of pleurobiliary fistula; therefore, the treatment plan should be tailored to individual patient needs. Endoscopic manoeuvres to relieve biliary obstruction and achieve drainage can obviate the need for high-risk surgery. Initial treatment is conservative in the form of intercostals drainage, drainage of any other collections, rehydration and correction of any metabolic abnormalities and subcutaneous administration of octreotide. Enteral nutrition and broad-spectrum antibiotics are very helpfully. Singh et al. [14], recommend endoscopic sphincterotomy to relieve the symptoms within 72 to 96 hrs after intercostals tube insertion and percutaneous drainage of the collection [18].

Gugenheim et al. [5], recommended external biliary drainage by percutaneous or surgical drainage of subphrenic abscess and/or direct percutaneous drainage of the intrahepatic biliary radices and treatment of the underlying aetiology. With biliary obstruction, the priority management was to treat the biliary disease [5,16,22].

Rapid drainage of bilious pleural effusion, maximum biliary decompression, prophylactic antibiotics, and the possible use of somatostatin have been recommended as initial treatments [16].

Chest tube and or pigtail were inserted in all patients as diagnostic and therapeutic. Greenish effusion of a large amount was the key and red flag to diagnose biliothorax. US abdomen and or CT chest abdomen were performed to detect any biliary dilatation, collection and or bile leak were performed once the diagnosis was reached or suspected. Conservative treatment in the form of chest tube or pigtail drainage of the biliothorax, fluid and electrolyte replacements and bed rest was needed in 28 cases (77%), ERCP intervention with or without percutaneous drainage of the liver or subphrenic abscesses. ERCP dilates the stenotic or obstructed hepatobiliary tracts with or without stents and or sphincterotomy. Surgical intervention was needed in 18 cases; 11 laparotomies and 7 thoracotomies. Two patients needed laparotomy followed by thoracotomy few weeks later. Fourteen patients needed more than one line of treatment. We started with chest tube which was successfully in 4 cases only. Then according to the CT, MRCP and ERCP, the line of further management was followed. Five cases needed chest tube insertion followed by laparotomy followed by thoracotomy. During our study, 4 patients died (11%), three from advanced malignancy and one from severe malnutrition and debilitation. One patient developed pleuro-bilio-cutaneous fistula and one developed entero-bilio-cutaneous fistula. Liao et al. [16], reported mortality rate of 8.8% and failure rate of 11.7%. They defined the treatment failure as persistence of symptom and or death due to bronchobiliary fistula or complications from the treatment. The prognosis is severe with an elevated rate of mortality of Moumen and el Fares reported a mortality rate of 12.2% [16,23,24].

Conclusion

Biliothorax is a presentation of a major pathology which needs a multidisciplinary treatment and staged prolonged management.

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References

- Crnjac A, Pivec V, Ivanecz A. Thoracobiliary fistulas: Literature review and a case report of fistula closure with omentum majus. *Radiol Oncol.* 2013;47(1):77-85.
- Ferguson TB, Burford TH. Pleurobiliary and bronchobiliary fistulas. *Arch Surg.* 1967;95(3):380-6.
- Warren KW, Christophi C, Armendariz R, Basu S. Surgical treatment of bronchobiliary fistulas. *Surg Gynecol Obstet.* 1983;157(4):351-6.
- Gandhi N, Kent T, Kaban JM, Stone M, Teperman S, Simon R. Bronchobiliary fistulas after penetrating thoracoabdominal trauma: Case report and literature review. *J Trauma.* 2009;67(5):143-5.
- Gugenheim J, Ciardullo M, Traynor O, Bismuth H. Bronchobiliary fistulas in adults. *Ann Surg.* 1988;207(1):90-4.
- Eryigit H, Oztas S, Urek S, Olgac G, Kurutepe H, Kutlu CA. Management of acquired bronchobiliary fistula: 3 case reports and literature review. *J Cardiothorac Surg.* 2007;2:52.
- Al-Qahtani HH. Biliopleural fistula with choles thorax. A rare complication of percutaneous transhepatic biliary drainage. *Saudi Med J.* 2011;32(11):1189-92.

8. Truong V, Huaranga A. Infected bile in the bellows: A case of pyocholethorax. *The Internet Journal of Pulmonary Medicine*. 2013;15:1.
9. Amir-Jahed AK, Sadrieh M, Farpour A. Thoracobilia: A surgical complication of hepatic echinococcosis and amebiasis. *Ann Thorac Surg*. 1972;14(12):198-205.
10. Kolbakir F, Erk MK, Keceligil HT, Yilman M. Cholethorax. *J Exp Clin Med*. 1994;11(1):45-46.
11. Córdoba López A, Monterrubio villar J, Bueno alvarezarenas. Biliothorax unrelated to fistula: A rare complication in gallbladder disease. *Arch Bronconeumol*. 2008;44(7):396-7.
12. Light RW, Gary LEE YC, Heffner JE. *Textbook of pleural Diseases*. 3rd ed. California, USA: CRC Press; 2016.
13. Pisani RJ, Zeller FA. Bilioid pleural effusion following liver biopsy. *Chest*. 1990;98(6):1535-7.
14. Singh B, Moodley J, Sheik-Gafoor MH, Dhooma N, Reddi A. Conservative management of thoracobiliary fistula. *Ann Thorac Surg*. 2002;73:1088-91.
15. Prabhu R, Bavare C, Purandare H, Supe A. Pleuro-biliary fistula-- a delayed complication following open cholecystectomy. *Indian J Gastroenterol*. 2005;24(1):28-9.
16. Liao GQ, Wang H, Zhu GY, Zhu KB, Lv FX, Tai S. Management of acquired bronchobiliary fistula: A systematic literature review of 68 cases published in 30 years. *World J Gastroenterol*. 2011;17(33):3842-9.
17. Ragozzino A, De Rosa R, Galdiero R, Maio A, Manes G. Bronchobiliary fistula evaluated with magnetic resonance imaging. *Acta Radiol*. 2005;46(5):452-4.
18. Karabulut N, Cakmak V and Kiter G. Confident diagnosis of bronchobiliary fistula using contrast-enhanced magnetic resonance cholangiography. *Korean J Radiol*. 2010;11(4):493-6.
19. Frampton AE, Williams A, Wilkerson PM, Paterson IM. Thoracobilia: A rare complication of gallstone disease. *Ann R Coll Surg Engl*. 2010;92(5):W1-3.
20. Jain SK, Gupta A, Kaza RC. Pleurobiliary fistula secondary to choledocholithiasis - A rare entity. *Asian J Surg*. 2008;31:29-31.
21. Artunduaga M, Patel NR, Wendt JA, Guy ES, Nachiappan AC. Bronchobiliary fistula localized by cholescintigraphy with single photon emission computed tomography. *World J Nucl Med*. 2015;14(1):60-2.
22. Rose DM, Rose AT, Chapman WC, Wright JK, Lopez RR, Pinson CW. Management of bronchobiliary fistula as a late complication of hepatic resection. *Am Surg*. 1998;64(9):873-6.
23. Mohammed M, Kobayashi K, Jawed M. Biliary pleural fistula following portal vein embolization for perihilar cholangiocarcinoma. *Case Rep Gastroenterol*. 2017;11(2):277-83.
24. Moumen M, el Fares F. Bilio-bronchial fistula of hydatid origin. Apropos of 8 cases. *J Chir (Paris)*. 1991;128(4):188-92.