Bronchobiliary fistulae due to echinococcosis

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Abstract

Objective: A bronchobiliary fistula (BBF) is a rare complication of echinococcosis due to rupture of hydatid cysts located at the upper surface of the liver to the bronchial tree. We present our experience in treating this uncommon and dangerous entity. Material: During the last 20 years, 21 patients, ten men and 11 women ranging in age from 26 to 83 years with a BBF were treated in our department. They presented dyspnea, biloptyysis, cough or fever. Diagnostic imaging studies have been very helpful in identifying the communication and in delineating its location. The disease was limited to the liver in 11 cases, whereas in the rest ten cases, both liver and lung were involved. Results: Right thoracotomy was the approach of choice. Our strategy consisted of adequate evacuation of the intrahepatic cysts, obliteration of the cyst space, freeing the adherent lung, dissection and closure of the BBF. Two deaths occurred due to anaphylactic shock and cardiac insufficiency. Follow up at 7–12 years did not reveal any recurrence. Conclusion: Although the incidence of echinococcosis has been decreased, the BBF still remains a serious complication with a high morbidity and mortality. Early diagnosis and management of septic associated complications are essential. © 2002 Elsevier Science B.V. All rights reserved.

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1. Introduction

A broncho-biliary fistula (BBF) is defined as an abnormal communication between the biliary system and the bronchial tree due to trauma, hemihepatectomy and necrotic hepatic infections, such as hydatid disease [1–3].

We present our experience in treating this rare entity produced by rupture of hydatid cysts of the liver dome to the bronchial tree.

2. Material and methods

From 1979 through 1999, 21 patients with a BBF were admitted at the Thoracic Surgery Department of General Hospital of Nikea – Piraeus. We retrospectively reviewed all patients’ records and analyzed surgical treatment and outcome. There were ten men and 11 women ranging in age between 26 and 83 years. All of them were infected by echinococcus granulosus.

2.1. Clinical presentation and physical examination

The main symptoms the patients complained for were dyspnea, biloptyisis, pleuritic chest pain, cough and high fever. Most of them had increased sputum production varying in appearance from clear to yellow purulent expectoration and ranging in quantity between 100 and 450 ml/day. Others experienced symptoms such as night sweats, anorexia and general fatigue. On physical examination decreased breath sounds were found over the right basal lung fields and hepatomegaly was present in 11 patients.

2.2. Diagnostic tools

2.2.1. Radiology

Chest X-ray and bronchography from 1975 to 1985, and liver sonography and CT scan from 1986 to 1999 have been very helpful diagnostic tools. Radiographic findings usually included a pleural effusion or an atelectasis of the lower pulmonary lobe associated in 18 cases with an elevation of the right hemidiaphragm. Air-fluid level suggesting abscess formation was also seen. At the beginning of our series bronchography was performed as a routine examination by the thoracic surgeons transcricoidally in the Radiology Department and it turned to be 100% diagnostic (n = 15) (Fig. 1). After the introduction of liver sonography and computed tomographic scan, as a routine, bronchogra-
phy was abandoned. A BBF was radiologically suspected when there was liver echinococcosis at the dome of the liver extending to the diaphragm. The sections passed through the lower thorax followed by subsegmental atelectasis of the middle or lower lobe and parenchymal infiltration of the basal segments.

2.2.2. Bronchoscopy

Bronchoscopy had a limited role because it was impossible to visualize the fistula due to the brisk flow of bile and the small size of the tract. It was mainly used for bronchial hygiene.

2.2.3. Laboratory tests

Among the laboratory tests only indirect hemagglutination test and immunofluorescence turned to be useful. Sputum analysis was routinely performed in order to identify possible viable scolices or membranes, since their presence in sputum is highly diagnostic in both BBF and rupture of a sole lung cyst. Bilirubin in the sputum was a clear laboratory evidence in about 40% of our cases and established the diagnosis.

2.3. Location of the fistula

In 11 cases the disease was limited to the liver, whether in the rest ten cases, both liver and lung were involved. Right lower lobe was usually involved, while one patient presented secondary involvement of the right pleural space.

3. Results

3.1. Surgical treatment

All but one, who denied operation, were treated surgically. Two of our patients had undergone previous operations for lung hydatidosis. A transthoracic approach was employed in all our cases through a right standard posterolateral thoracotomy. Dissection of the adherences to the lung has been the first step. Through a right thoracophrenotomy the dome of the liver was exposed, saline pads were used to protect from spillage, the BBF was found and dissected, and the debridement of the liver cyst began. After the total evacuation of the pericyst a thick tube drainage was placed subdiaphragmatically. The thoracophrenotomy was sutured and the lung lesions had to be confronted. The necrotic lung tissues were removed from the thoracic cavity trying to save as much as possible healthy lung tissue and after the insertion of two chest tubes the thoracotomy was sutured in layers. In all our cases we kept the drainage of the liver cyst in place until there was clear evidence for its reduction. We supervised the healing process through radio-

Fig. 1. Bronchography showing a communication of the thoracic and abdominal cavities.
graphic control of the cavity every week, and when there was a satisfactory reduction we removed the drainage.

3.2. Morbidity – mortality

Prolonged air leak, atelectasis were the major complications that occurred in two patients. All were conservatively treated. The 30-day mortality was 9.5%. Two of our patients died postoperatively, one man of 55 years due to severe cardiac insufficiency and one woman of 50 years due to non-reversible anaphylactic shock. The last patient was admitted to the emergency department in shock, which was caused by the rupture of a cyst in the pleural cavity.

3.3. Follow up

Follow up at 7–12 years revealed no recurrence. Five of our patients received postoperatively medical treatment with albendazole for a period of 6 months.

4. Comments

One of the main complications of the hydatid cyst of the dome of the liver is its rupture into the bronchial tree [3,4]. This 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 [3,5]. When the cyst crosses the diaphragm, the possibility of rupture into the pleural cavity, or even into great thoracic vessels [6] or pericardium increases [6]. Allergic reactions in these cases are common and potentially lethal [3,4], as it was the case in one of our patients.

Although in many series [7,8] bronchography is not considered to be a useful diagnostic tool, in our patients when performed, it easily demonstrated the fistula and also revealed bronchiectatic lesions, as it occurred in one of our patients. Today there are less invasive methods such as computed tomographic scan, ultrasonography and nuclear imaging that turned to be very important diagnostic tools [7,8]. The information they provide about the local extension of the disease, the existence of other cysts in adjacent organs and the identification of a BBF are essential to the surgeon. Taking also in consideration that the recurrence rate is high, these non-invasive methods are ideal for follow up and early detection of complications such as BBF, rupture or other life-threatening conditions.

Once a BBF develops, cure is possible only with correction of possible biliary obstruction, adequate drainage of any concomitant abscess and of course treatment of the underlying disease [9–11]. Endoscopic treatment of BBF has been reported only as a first stage treatment until the patient’s condition gets stable [12]. Surgery still remains the treatment of choice in case of echinococcosis [10,11]. The approach of choice in our series was a right standard posterolateral thoracotomy because of the facility that offers in exposing both lung and liver. Extended pulmonary resections are not justified taking into account that echinococcosis has always the risk of recurrence and there is a vital need for sparing as much viable lung tissue as possible [10–14]. Only in rare cases with necrotic inflammatory lung tissue, diffusely involved, segmentectomy or even lobectomy can be carried out [3,11,15]. In our cases there was no need for such kind of operations.

Medical treatment with mebendazole or albendazole has been reported [7,10,12]. It is mainly used perioperatively in cases of liver hydatidosis with very small cysts, particularly in children and in cases of diffuse disease with uncertain results [10].

In conclusion, although the incidence of hydatidosis has been really decreased today rupture of hydatid cysts to the bronchial tree still remains a very dangerous complication with a high morbidity and mortality. Careful assessment and early treatment of septic complications are essential in successfully treating this rare condition.

References