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Original Article

Abdominothoracic fistulas due to complicated Echinococcosis: surgical treatment and outcome

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ABSTRACT

Background: Abdominothoracic fistulas are severe complications of hydatid disease. We report here on the results of treatment of hydatid abdominothoracic fistulas in 17 patients.

Material and Methods: A single-center case series of 17 patients with abdominothoracic fistulas (ATF) was treated at our hospital from 2004 and 2019. Nine men and eight women patients (age range: 46-85 years; median age: 59 years) were treated for abdominothoracic fistulas (ATF). The main symptoms were dyspnea, chest pain, cough, purulent sputum, high fever in 15 patients and additionally biliptysis in two patients. Fistulas were hepatopleural (HPF) in 14, bronchobiliary (BBF) in two and abdominopleural in one patient.

Results: Fourteen patients were operated for HPF and 3 (18%) patients were treated with percutaneous transhepatic drainage and tube thoracostomy. All patients were discharged from the hospital in good health. Our strategy consisted of adequate evacuation of the intrahepatic cyst,closure of the fistula via thoracophrenotomy and long term drainage of the intrahepatic or subhepatic cyst space up to ceased biliary drainage.

Conclusions: ATF due to hydatid cyst is uncommon. In rare cases ATF may be present at the abdominal,thoracic or diaphragmatic level. Thoracophrenotomy is the best surgical treatment for all three levels.In unstable patients only long-term percutaneous drainage should be applied. Medical treatment with Albendazole is indicated when dissemination is confirmed. Early diagnosis and management of septic associated complications are main goal.

Keywords: abdominopleural fistula, complication, cyst hydatid

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Introduction

An abdominothoracic fistula (ATF) is defined as an abnormal communication between the abdomen and the bronchial tree or pleura due to trauma or hydatid or amoebic liver disease [1]. Although a rare entity, it is considered a severe complication of hydatid liver disease. A liver cyst may rupture into the biliary tree, a hollow organ, through the diaphragm into the pleural cavity, or directly into the peritoneal cavity [2,3]. Intrapulmonary rupture of a hepatic hydatid cyst is usually uncommon and the underlying cause is mostly due to the hepatic hydatid cyst perforation to the right subphrenic area and then fistulation into the posterior basal segment of the right lower lobe of the lung [4]. Management of these fistulas is often very difficult and can be associated with high morbidity. Surgical closure of the fistula tract with efficient drainage of the chest and cystic cavity is the first and the most common treatment approach [5]. There exists no clear consensus or guidelines for the management of life-threatening complications.

We report here on our practice of treating 17 patients with ATF.

Materials and Methods

We retrospectively reviewed the patient records of 17 patients with ATF due to hydatid disease who had been treated between 2004 and 2019. Written informed consent was obtained from all patients; patients attested that they understood their treatment options, potential complications, expected risks and posssible benefits of treatment. The study was approved by the Institutional Ethics Committee (Reference No:TUTF-GOBAEK 2022/274). There were nine men and eight women, whose ages ranged between 46 and 85 years. A written informed consent was obtained from all patients. As a surgical approach, we performed drainage of the perforated cystic lesion via, thoracophrenotomy, preferring partial pericystectomy to liver resection because of the need to avoid the redundant loss of normal parenchyma. Cyst aspiration before removal was the essential method of treatment for intact cysts of liver. The purulent contents of the hepatic and pleural cavity were cleared, and it was irrigated intraoperatively with povidone-iodine or hypertonic saline used as scolicidal agents. The area of fibrous tissue and abscess on the diaphragm was debrided; the fistula tract was closed, and segment or lob of involved lung was resected (Figure 1A). A subdiaphragmatic drain was placed in all patients undergoing thoracophrenotomy, and it was left in place for a few days or until the output bile drainage was minimal (which ranged from 5 days to 3 months) in patients with bile drainage. Endoscopic retrograde cholangiopancreatography (ERCP) had been carried out in two patients by the gastroenterologist for continued leakage of bile from PTHD before surgery for biliary decompression. Bile leakage decreased but did not stop after ERCP; therefore, both underwent surgical treatment. There was no need for biliary decompression (ERCP) postoperatively. Postoperatively, albendazole 800 mg/d was given to all patients during 3 months to prevent recurrence. The follow-up period included monthly biochemical tests for liver function and clinical examination in patients using albendazole. After discharge, radiologic examination was applied in the third and sixth months and in the first year with chest x-ray and thorax CT if required.

Results

The main symptoms were cough, dyspnea, chest pain and purulent sputum in 15 (88%) patients and biliptysis in two patients. Six patients underwent previous operation or intervention for liver hydatid disease. The patients with biliptysis had increased bile-stained expectoration that ranged from 100 and 500 mL/day in quantity. On physical examination, decreased breath sounds were found over the right basal lung areas in all patients, except one. The diagnostic work-up included biochemical tests for liver function, chest x-rays and computed tomography, which were applied routinely to detect hepatic and pulmonary cysts. A fistulous tract reaching to the basal segments of the right lung was shown with contrast injection through the catheter inserted under fluoroscopic guidance into the hepatic cystic cavity in the two patients with biliptysis (Figure 1B). Thorax computed tomography (CT) revealed right lower lobe pneumonia, pleural thickening or destroyed basal segments of the right lower lobe in 10 (59%) patients (Figure 2). Four patients were also cysts with concomitant liver and right lower lobe. Air-fluid level suggesting subdiaphragmatic abscess formation was also seen most of the patients. However, it was often difficult to demonstrate the fistula itself before surgery. In the sputum and pleural fluid analysis, viable scolices or membranes were revealed in 10 (59%) patients. The results of diagnostic evaluation demonstrated that fistulas were hepatopleural (HPF) in 14 (82%) patients, bronchobili-

ary (BBF) in two and abdominopleural (APF) in one. The predisposing pathology is summarized in Table 1. No scolicidal drugs were used preoperatively. Serological and skin testing for hydatid disease were not part of our routine diagnostic tests. Two patients with biliptysis were started on a diet including low fat. First, A percutaneous transhepatic drainage (PTHD) into the cyst cavity had been performed by interventional radiologists in six patients, and symptomatic healing of the abdominal symptoms but not of the biliptysis was observed. In three of these patients, only concomitant drainages of both PTHD and TT were performed, due to the patient's generally poor condition. In seven patients; tube thoracostomy (TT) was first performed because they were referred with pleural fluid accumulation. Surgical treatment is summarized in Table 2. Twelve patients underwent right thoracophrenotomy in the seventh or eighth intercostal space for the destruction of lung parenchyma due to biliary destruction, prolonged compression or infection. Right lower decortication was performed with video assisted thoracoscopic (VATS) in one patient. A left thoracophrenotomy was applied in one patient due to a parenchyma left lower lobe related to abdominopleural fistula. We performed lobectomy in two patients due to destroyed lobes and wedge resection in six patients for the damaged lung part. There was extensive evidence of the thickened pleura with empyema intraoperatively in four patients, and all of them underwent decortication. Cystotomy and capitonnage were performed for the cyst localized to the right lower lobe in four patients. All patients were discharged in good condition. There was no 30-day mortality in our series. The treatment algorithm we used in patients with ATF based upon the our experience is summarized in Figure 3.

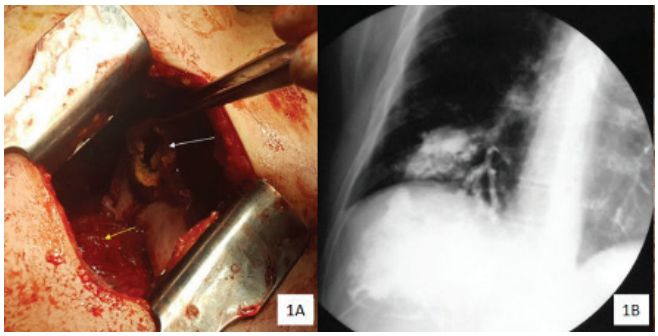

Figure 1. Peroperative view shows fistula tract in diaphragm (white arrow) and destroyed lung tissue (yellow arrow) **(A)**, fistulogram revealed a broncho-biliary fistula on the liver between bronchial tree **(B)**.

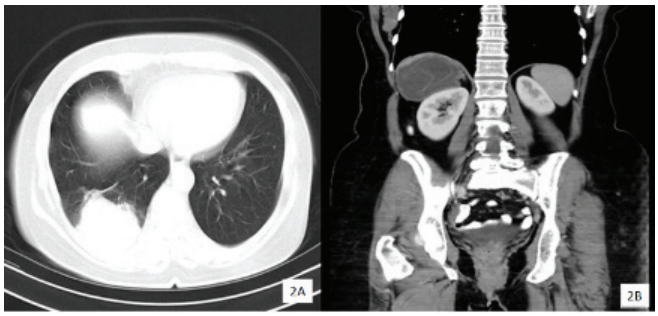

Figure 2. Computed tomography demonstrating consolidation of the lateral basal segment of the right lobe **(A)**, boronal section computed tomography same patient showing a right subdiaphragmatic cystic cavity **(B)**.

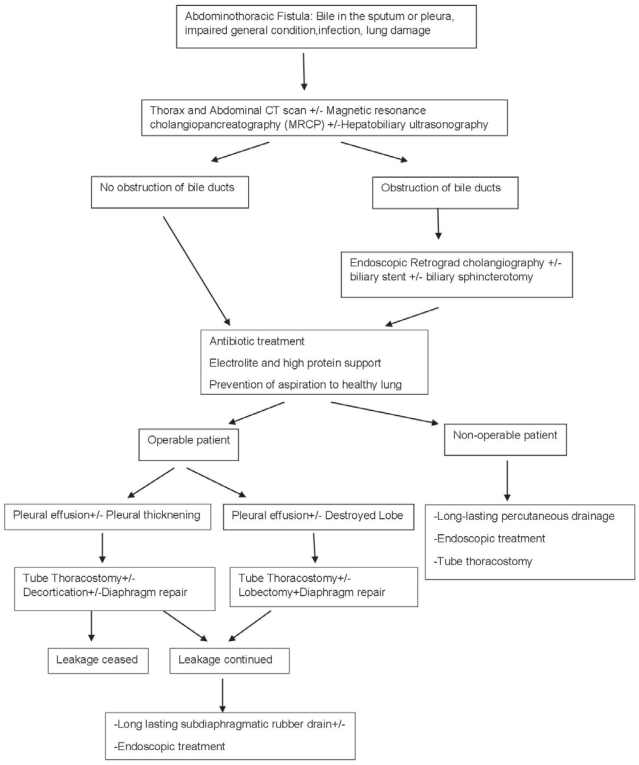

Figure 3. Algoritm for ruptured hydatid cyst in the thorax with biliobronchial fistula.

Discussion

Cyst hydatid disease remains a serious sanitary problem for the Middle East, Australia, New Zealand, and Mediterranean countries, including Turkey. Abdominothoracic fistula carries a high mortality risk of 10.3% according to the literature, mainly due to surgical complications [6]. Abdominal cyst hydatid rupture and erosion of the diaphragm lead to passage of the contents of the cyst through the diaphragm into the pleural space, parenchyma or bronchus, causing pneumonitis, empyema and bronchobiliary fistula (BBF). Abdominothoracic fistula can occur in several ways: hepatopleural (HPF), bronchobiliary (BBF) or abdominopleural fis-

Table 1. Patients' demographic data, including fistula type, pathology, and the presence of bile and scolices in body fluids.

No	Age	Sex	Fistula Type	Bile	Scolex	Pathology
1	62	F	APF	Ø	Ø	Cystic abscess in splenic zone
2	62	M	BBF	Sputum	Sputum	Liver and lung cyst
3	47	M	BBF	Sputum	Ø	Liver cystic abscess
4	59	M	HPF	Pleural fluid	Ø	Liver cyst
5	53	F	HPF	Ø	Pleural fluid	Liver cyst
6	60	F	HPF	Pleural fluid	Pleural fluid	Liver cyst
7	73	F	HPF	Pleural fluid	Pleural fluid	Liver and lung cyst
8	46	M	HPF	Pleural fluid	Pleural fluid	Liver and lung cyst
9	70	M	HPF	Pleural fluid	Pleural fluid	Liver cyst
10	54	F	HPF	Pleural fluid	Pleural fluid	Liver cyst
11	65	M	HPF	Pleural fluid	Pleural fluid	Liver cyst
12	52	F	HPF	Pleural fluid	Ø	Liver cystic abscess
13	56	F	HPF	Pleural fluid	Ø	Liver and lung cyst
14	58	M	HPF	Pleural fluid	Pleural fluid	Liver cyst
15	72	M	HPF	Pleural fluid	Ø	Liver cyst
16	29	F	HPF	Pleural fluid	Pleural fluid	Liver cyst
17	85	M	HPF	Ø	Ø	Liver cyst

Abbrev.: APF: abdominopleural fistula; BBF: bronchopleural fistula; HPF: hepatopleural fistula.

tula (APF). Clinical features and symptoms preceding the development of ATF can be distinctive according to fistula type. APF causes irritating cough, chest pain and pleural effusion and basilar atelectasis. HPF includes fever, chest pain, productive cough, right upper abdominal pain, presence of bile in the pleural fluid, sometimes jaundice. BBF characterized bile stained sputum, fever, findings of sepsis and poor general status [7,8] (Table 1). Unstable septic patients in admission need hemodynamic, respiratory and nutritional support, followed by as early as surgical intervention.

Diagnosis can be radiological or include interventional methods. Thorax CT can reveal hepatic cyst and abscess, and pleural effusion; however, it does not show fistula tract in most patients. If bile-stained sputum or pleural fluid or jaundice detected, a more precise examination is required. In this situation, magnetic resonance imaging (MRI) and MR cholangiography can be useful tools, although there are publications that suggest that it can also fail to indicate the fistulous tract [9]. If a fistula is suspected and there is jaundice, this may indicate the need for further investigation and treatment using ERCP or percutaneous transhepatic cholangiography (PTC) for fistula and possible distal biliary obstruction [10-12]. There had been continued leakage of bile whereupon ERCP had been applied by the gastroenterologist; in two patients with BBF, however, leakage continued. Upon this, we performed liver cystotomy with right thoracophrenotomy and subdiaphragmatic drain inserted; therefore, leakage ceased after 1 and 3 months in two patients. Some authors have indicated cases of spontaneously closure of the fistula after ERCP or PTC. Other authors have stated that the more conservative methods require a long-lasting drainage period as long as 5 weeks [6,13]. If chronic fistulas associated with thoracic and abdominal sepsis and respiratory impairment is present, surgery should be considered for such patients as the gold standard.

Table 2. Surgical treatment options for abdominothoracic fistulas.

No	First attempt	Surgery
1	Ø	Left lower lobectomy
2	PTHD+ERCP	Cystotomy,capitonnage, Liver cystotomy
3	PTHD+ERCP	Liver cystotomy, tractotomy
4	TT	RLL decortication, Liver cystotomy
5	PTHD	TT
6	TT	RLL wedge, Liver cystotomy
7	TT	RLL wedge, Liver cystotomy
8	Ø	RLL wedge, Liver cystotomy
9	Ø	RLL lobectomy
10	PTHD	RLL wedge, Liver cystotomy
11	PTHD	TT
12	TT	VATS decortication
13	TT	RLL cystotomy, capitonnage decortication, Liver cystotomy
14	Ø	RLL wedge, Liver cystotomy
15	TT	RLL wedge, Liver cystotomy
16	TT	RLL decortication, Liver cystotomy
17	PTHD	TT

Abbrev.; Endoscopic retrograde cholangiopancreatography (ERCP), PTHD: percutaneous transhepatic drainage; RLL: right lower lobe; TT: tube thoracostomy.

Once ATF develops, a cure is possible only with the removal of possible biliary obstruction, efficient drainage of any associated abscess and treatment of the underlying disease [1]. Drainage of pleural infection, excision or decortication of destroyed lung tissue and fistula tract through phrenotomy should be performed. Moreover, biloma, abscesses and any other pathology in the subdiaphragmatic area can be cleared and a long-lasting rubber drain can be inserted. Ferguson and Burford [14] reported that successful management of the fistula with immediately aggressive surgery thoracotomy, efficient subdiaphragmatic drainage using phrenotomy, careful closure of the diaphragmatic fistula tract with non-absorbable sutures, decortication of a thickened pleura. Standard thoracotomy has been offered as a better approach to BBF because it provides radical treatment of bronchopulmonary disease [15-17]. Tocci et al [18] claimed that standard thoracotomy was unsafe due to lack of control of the hepatic pedicle and access to the biliary tree; therefore, it may result in serious intraoperative risks. In their study, they recommended thoracoabdominal incision for good access to the hepatic pedicle and to the biliary tract to enable hemostatic maneuvers and any needed surgical intervention on the biliary tract if needed. The approach of an option in our patients was a right thoracotomy and phrenotomy through the seventh or eighth intercostal space, as this proposed good exposure to the subphrenic abscess and of the destroyed lung tissue. Radical surgical approaches (e.g., resection) were performed in case of destruction of the enclosing lung parenchyma or after cyst removal, re-expansion problem of atelectatic lung. Six wedge resections and two lobectomies were performed due to the destroyed or compressed lung parenchyma in our series. In four patients of empyema and due to thickened pleura, lung decortication was completed the surgical approach. In patients with coexisting abdominal cyst, thoracophrenotomy is the best procedure for surgical treatment at all three levels. As we mentioned earlier, according to previous publications, subcostal incision and thoracophrenotomy during one stage operation provided a larger surgical field above and below the diaphragm [5]. The most difficult step of the operation may be the repair of inflamed diaphragms. Large defects can be managed with mobilization of nearby tissue (pericardial fat, intercostal muscle, omentum) or synthetic mesh. However, in most cases, primary closure with non absorbable sutures to close the defect was enough [6,19]. None of our patients required any other methods except primary suture.

There was no mortality and morbidity as a result of the one-stage operation. In case of the concomitant hepatic hydatid cyst below the diaphragm, removal of both liver and lung lesions during one stage operation is the best option. Rabiou et al [20] proposed that exclusive endoscopic sphincterotomy may be an alternative in some non-operable patients. In our two patients, bile leakage continued despite sphincterotomy PTHD. These patients were operated. Bile drainage was interrupted only with PTHD in three of our patients who were not suitable for the operation. We believe that medical treatment after surgery is required for a ruptured intrathoracic hepatic hydatid cyst, when there is dissemination or when total evacuation of the cyst is impossible. We recommend albendazole 400 mg twice daily for 3 to 6 months in such patients. Kuzucu et al [21] also recommended daily treatment with albendazole 800 mg for at least 2 months after surgery for all patients with multiple hydatid or complicated cysts or both.

In conclusion, although the frequency of cyst hydatid disease has decreased, rupture of hydatid cysts to the bronchial tree and pleura still remained as a severe complication that can lead to high morbidity and mortality. Careful evaluation and early surgical treatment without delay and before the development of septic complications are the main requirements. We believe that the thoracophrenotomy procedure is a good and safe choice of surgical treatment at all three levels. The patients with poor general condition, only long-lasting percutaneous drainage should be performed. Medical treatment with albendazole is recommended when dissemination is approved. This study is a retrospective non-randomized small sized patient population and single center study. However, further long-term studies including more patients and longer follow-up intervals are required

Declaration of conflicting interests

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Ethics approval

The study was approved by the Instutional Ethics Committee (Reference No:TUTF-GOBAEK 2022/274).

Authors' contribution

YAK: conceptualization, methodology, design of the work, formal analysis, acquisition writing-orginal draft FY: investigation, data curation, YY: supervision.

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