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Abdominothoracic Fistulas Due To Complicated Echinococcosis

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ABSTRACT

Background: Abdominothoracic fistulas are serious complications associated with hydatid disease. We present the results of treating hydatid-related abdominothoracic fistulas in a cohort of 19 patients.

Materials and Methods: This study is a single-center case series involving 19 patients treated for abdominothoracic fistulas (ATF) at our hospital between 2004 and 2023. The group comprised ten men and nine women, with an age range of 46 to 85 years and a median age of 59 years. The primary symptoms observed included dyspnea, chest pain, cough, purulent sputum, high fever in 17 patients, and biliptysis in two patients. Among the fistulas, 16 were classified as hepatopleural (HPF), two as bronchobiliary (BBF), and one as abdominopleural (APF).

Results: Sixteen patients successfully underwent surgical interventions for HPF, while three patients (18%) were effectively managed with percutaneous transhepatic drainage and tube thoracostomy. All patients were discharged in good health, demonstrating the effectiveness of our treatment strategy. Our approach involved thorough evacuation of the intrahepatic cyst, closure of the fistula via thoracophrenotomy, and long-term drainage of the intrahepatic or subhepatic cyst space until biliary drainage ceased.

Conclusions: Although ATF resulting from hydatid cysts is uncommon, early diagnosis is critical when it does occur. In rare instances, ATFs may manifest at the abdominal, thoracic, or diaphragmatic levels. Thoracophrenotomy is considered the optimal surgical intervention for all three levels. In unstable patients, long-term percutaneous drainage is advised. Medical treatment with Albendazole is warranted when dissemination is confirmed. Our primary objectives are the early diagnosis and management of septic complications, highlighting the essential role of medical professionals in the timely and effective treatment of ATF.

Key Words: Abdominopleural fistula, complication, cyst hydatid

Introduction

An abdominothoracic fistula (ATF) is a rare and complex condition characterized by an abnormal connection between the abdomen and the bronchial tree or pleura, often resulting from trauma or associated with hydatid or amoebic liver disease [1]. Despite its rarity, ATF is recognized as a severe complication of hydatid liver disease. A liver cyst may rupture into the biliary tree, a hollow organ, penetrate through the diaphragm into the pleural cavity, or directly invade the peritoneal cavity [2,3]. Intrapulmonary rupture

of a hepatic hydatid cyst is infrequent. It typically occurs when the cyst perforates the right subphrenic area, leading to a fistula into the posterior basal segment of the right lower lobe of the lung. Managing these fistulas presents considerable challenges and is often linked to significant morbidity. The first-line treatment involves surgically closing the fistula tract while ensuring adequate drainage of both the chest and the cystic cavity [5]. Currently, there are no established guidelines or consensus on managing these life-threatening complications. This report outlines our experiences in treating 17 patients with ATF.

Methodology:

We conducted a retrospective review of the medical records for 19 patients diagnosed with ATF caused by hydatid disease, treated between 2004 and 2024. All patients provided written informed consent, confirming their understanding of treatment options, potential complications, anticipated risks, and possible benefits. The Institutional Ethics Committee approved the study (Reference No: TUTF-GOBAEK 2022/274). Among the participants were ten males and nine females, aged 46 to 85 years. Written informed consent was obtained from each patient. The primary symptoms included cough, dyspnea, chest pain, and purulent sputum in 17 patients, while two patients experienced biliptysis. Six of the patients had undergone prior surgeries or interventions for liver hydatid disease. Those presenting with biliptysis reported bile-stained expectoration varying from 100 to 500 mL/day. All patients exhibited decreased breath sounds over the right lower lung areas during physical exams except one. Standard diagnostic evaluations included biochemical tests for liver function, chest X-rays, and computed tomography, routinely conducted to identify hepatic and pulmonary cysts. Findings from chest X-rays and thorax computed tomography (CT) indicated right lower lobe pneumonia, pleural thickening, or destruction of the basal segments of the right lower lobe in 10 patients (Fig. 1). Five patients also presented cysts affecting both the liver and the right lower lobe. Most patients showed signs of an air-fluid level, suggesting the presence of a subdiaphragmatic abscess. However, demonstrating the fistula before surgery was often challenging. In two patients with biliptysis, a fistulous tract connecting to the basal segments of the right lung was identified using contrast injection through a catheter inserted under fluoroscopic guidance into the hepatic cystic cavity. Sputum and pleural fluid analyses revealed viable scolices or membranes in 10 patients. The diagnostic evaluations indicated that fistulas were classified as hepatopleural (HPF) in 16 patients, bronchobiliary (BBF) in two, and abdominopleural (APF) in one. (Fig.2) The contributing pathology is summarized in Table 1. No scolicidal medications were administered preoperatively,

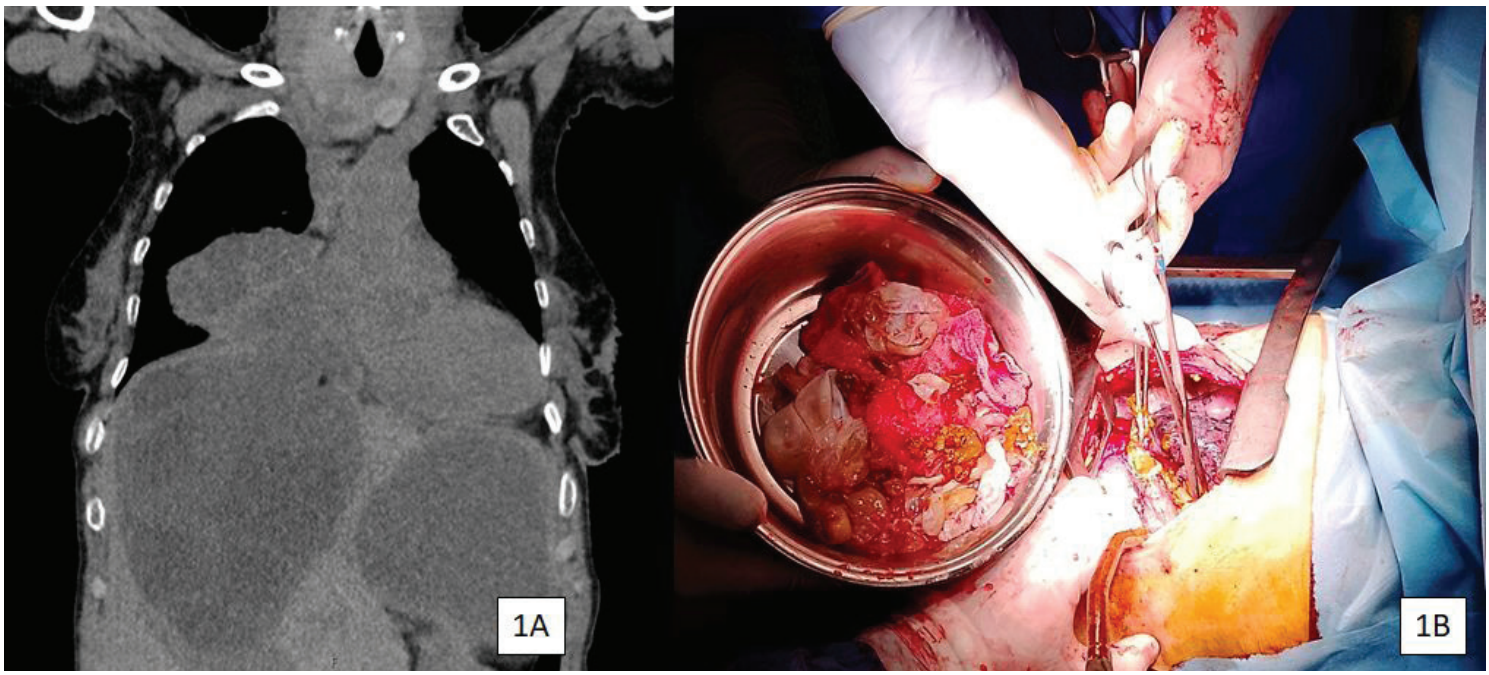


Figure 1.A Coronal section computed tomography demonstrating ruptured liver hydatid cyst in the right thorax.
1. B Intraoperative picture showing exposed contents and collapsed cyst wall

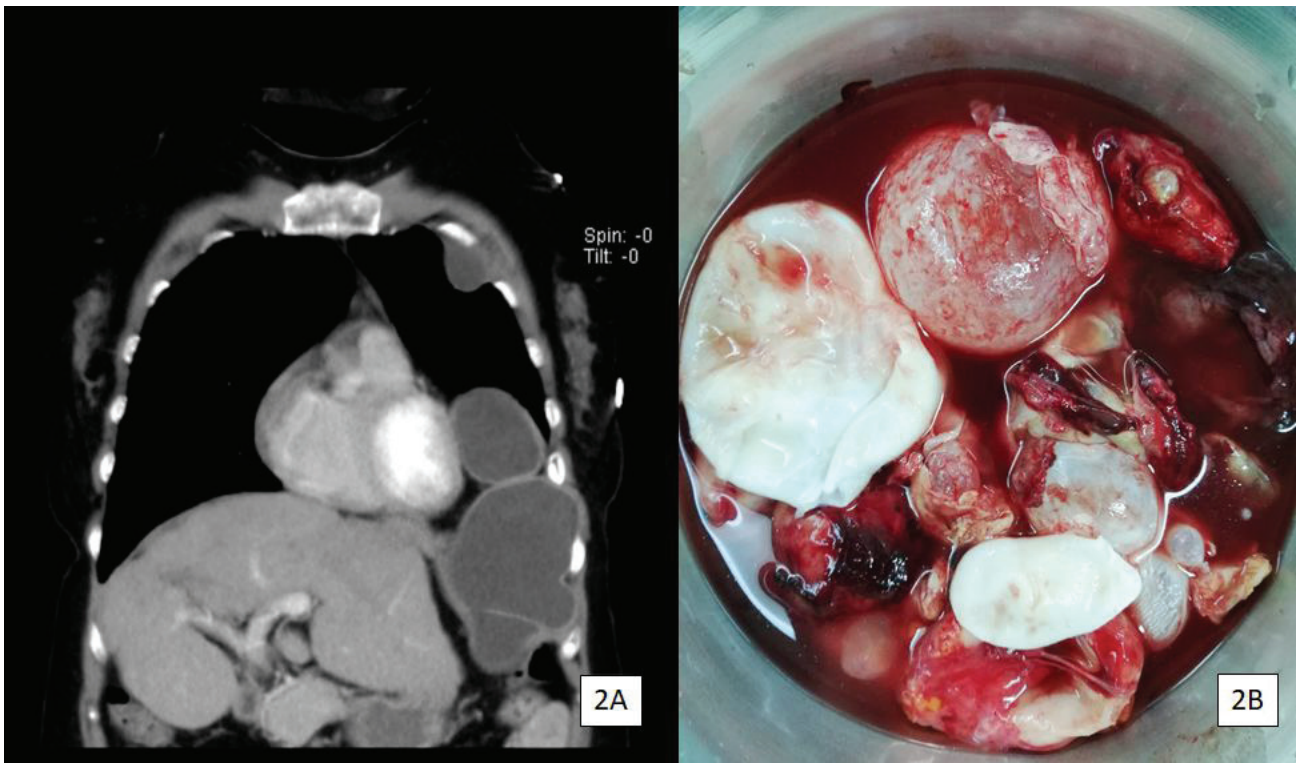


Figure 2. A Coronary section computed tomography shows that cyst hydatid originated from the subdiaphragmatic area in the left hemithorax**. 2.** B Extracted specimen of hydatid cyst with exposed contents.

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
18	50	M	HPF	Ø	Ø	Liver cyst
19	68	F	HPF	Ø	Ø	Liver and lung cyst

* APF: abdominopleural fistula; BBF: bronchopleural fistula; HPF: hepatopleural fistula

and serological and skin tests for hydatid disease were not included in our routine diagnostic procedures.

Results:

In this study, we documented two biliptysis patients placed on a low-fat diet. Before this intervention, six patients underwent percutaneous transhepatic drainage (PTHD) into the cyst cavity, leading to symptomatic relief of abdominal symptoms; however, there was no significant improvement in biliptysis. Among these, three patients received both PTHD and tube thoracostomy (TT) concurrently due to their generally poor clinical status. Seven patients required initial TT because they were referred with pleural effusions. Surgical interventions are detailed in Table 2. We performed right thoracophrenotomy via the seventh or eighth intercostal space in fourteen patients to address lung parenchyma damage secondary to biliary destruction, prolonged compression, or infections. In one case, right lower decortication was executed using video-assisted thoracoscopic surgery (VATS). One patient underwent left thoracophrenotomy due to abdominopleural fistula affecting the left lower lobe. Lobectomy was performed in two patients with destroyed lung lobes, while six patients underwent wedge resections for damaged lung sections. Upon intraoperative assessment, four patients presented with thickened pleura and empyema, all requiring decortication. For cysts localized in the right lower lobe, cystotomy and capitonnage were performed in three patients. During thoracophrenotomy, drainage of the perforated cystic lesion was prioritized, favoring partial pericystectomy over liver resection to minimize unnecessary loss of healthy parenchyma. Cyst aspiration before resection proved essential for intact liver cysts. Intraoperatively, purulent material was evacuated from the hepatic and pleural cavities, and irrigation was performed with povidone-iodine or hypertonic saline as scolicidal agents. The fibrous tissue and abscess on the diaphragm were debrided, and the fistula tract was closed, accompanied by segmental or lobar lung resection as necessary. Subdiaphragmatic drains were placed in all patients following thoracophrenotomy, remaining in situ for several days or until bile drainage became minimal, varying from 5 days to 3 months. Endoscopic retrograde cholangiopancreatography (ERCP) was performed in

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
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
18	Liver cystotomy+ERCP	RLL decortication, Liver cystotomy
19	Ø	RLL Cystotomy,capitonnage, Liver cystotomy

*Endoscopic retrograde cholangiopancreatography (ERCP), PTHD:Percutaneous transhepatic drainage, RLL: Right lower lobe, TT: tube thoracostomy.

two patients due to persistent bile leakage from PTHD before surgical intervention for biliary decompression, resulting in reduced but incomplete bile leakage. Consequently, both underwent surgical procedures.

All patients achieved good postoperative outcomes, with no postoperative biliary decompression via ERCP requirement and no 30-day mortality noted. Postoperatively, all patients were administered albendazole at 800 mg/day for three months to mitigate recurrence risk—follow-up involved monthly biochemical evaluations for liver function and clinical assessments for patients receiving albendazole. Radiological examinations, including chest X-rays and thoracic CT scans, were conducted at three, six, and twelve months post-discharge, as warranted. Notably, there was no in-hospital mortality in our cohort. An algorithm for managing ruptured hydatid cysts in the thorax with biliobronchial fistula, based on our clinical experience.

Discussion:

Cyst hydatid disease remains a significant public health concern in regions such as the Middle East, Australia, New Zealand, and Mediterranean countries, including Turkey. The occurrence of abdominothoracic fistula is associated with a notably high mortality rate of 10.3%, as reported in the literature, primarily due to surgical complications [6]. The rupture of abdominal cyst hydatid and the erosion of the diaphragm facilitates the migration of cyst contents into the pleural space, lung parenchyma, or bronchus, resulting in conditions such as pneumonitis, empyema, and broncho-biliary fistula (BBF). The abdominothoracic fistula may present in several forms: hepatopleural fistula (HPF), bronchobiliary fistula (BBF), or abdominopleural fistula (APF). The clinical features and symptoms preceding the development of ATF can vary significantly depending on the type of fistula. APF typically presents with a persistent cough, chest pain, pleural effusion, and atelectasis in the basal lung regions. HPF is characterized by fever, chest pain, a productive cough, pain in the right upper quadrant of the abdomen, bile within the pleural effusion, and occasionally jaundice. BBF presents with bile-stained sputum, fever, clinical signs of sepsis, and generally poor clinical status [7,8] (Table 1). Unstable septic patients, upon admission, necessitate comprehensive hemodynamic, respiratory, and nutritional support, followed by timely surgical intervention. Diagnostic evaluation may involve radiological imaging or interventional techniques. Thoracic computed tomography (CT) scans can reveal hepatic cysts, abscesses, and pleural effusions; however, they often do not visualize the fistula tract in most cases. In instances where bile-stained sputum pleural effusion or jaundice is identified, a more thorough examination is warranted. In such cases, magnetic resonance imaging (MRI) and MR cholangiography may be useful diagnostic tools, although some studies suggest they may also fail to delineate the fistulous tract [9]. Suppose a fistula is suspected and jaundice is present. In that case, this situation may require further investigation and therapeutic measures, including endoscopic retrograde cholangiopancreatography (ERCP) or percutaneous transhepatic cholangiography (PTC) to address the fistula and any potential distal biliary obstruction [10-12]. There have been instances of persistent bile leakage following ERCP performed by gastroenterologists; in two patients with BBF, leakage persisted. Consequently, we executed liver cystotomy along with right thoracophrenotomy and the insertion of a subdiaphragmatic drain; thus, the leakage subsided after 1 month and 3 months in the respective patients. Some researchers have reported cases where a fistula closed spontaneously after undergoing either Endoscopic Retrograde Cholangiopancreatography (ERCP) or Percutaneous Transhepatic Cholangiography (PTC) procedures. Conversely, other studies suggest conservative treatment options may require longer drainage times, potentially extending up to 5 weeks [13,14]. In patients with chronic fistulas associated with thoracic and abdominal infections and respiratory difficulties, surgical intervention should be considered the preferred method.

Once an abdominal-thoracic fistula develops, a definitive cure can only be achieved by removing any biliary blockage, effectively draining related abscesses, and properly treating the underlying condition. It is crucial to drain any pleural infections, excise or decorticate damaged lung tissue, and address the fistula tract through phrenotomy. Additionally, issues such as bilomas and abscesses in the subdiaphragmatic region should be treated, followed by the insertion of a long-lasting rubber drain. Ferguson and Burford [15] indicated that effective management of a fistula involves immediate aggressive surgical intervention through thoracotomy, thorough subdiaphragmatic drainage using phrenotomy, careful closure of the diaphragmatic fistula tract with nonabsorbable sutures, and decortication of thickened pleura. Standard thoracotomy has been proposed as a more advantageous method for treating bronchobiliary fistulas due to

its capability for radical treatment of bronchopulmonary diseases [16-18]. Tocci et al. [19] argued that standard thoracotomy raises safety concerns because it does not provide adequate control over the hepatic pedicle and access to the biliary system; consequently, this may lead to significant intraoperative complications. Their study advocated for a thoracoabdominal incision to ensure better access to the hepatic pedicle and biliary system, thus allowing for hemostatic techniques and necessary biliary surgical interventions. In our cases, a right thoracotomy combined with phrenotomy through the seventh or eighth intercostal space was chosen, as it provided optimal exposure of the subphrenic abscess and damaged lung tissue. Radical surgical methods, such as resections, were performed when there was significant destruction of the surrounding lung parenchyma or cyst removal was needed, followed by the re-expansion of atelectatic lung issues. Our cases included six wedge resections and two lobectomies due to compromised or destroyed lung parenchyma. For four patients suffering from empyema and associated thickened pleura, lung decortication was part of the surgical approach. The most effective surgical option for patients with concurrent abdominal cysts is thoracophrenotomy at all three levels. Earlier studies have shown that a subcostal incision combined with thoracophrenotomy in a single-stage surgery provides a more extensive surgical field both above and below the diaphragm [5]. One of the most challenging aspects of the procedure may involve repairing the inflamed diaphragms. Large defects can be closed by mobilizing adjacent tissues (such as pericardial fat, intercostal muscles, or omentum) or using synthetic mesh. However, in most cases, primary closure using nonabsorbable sutures was sufficient to seal the defect. None of our patients required any alternative methods apart from primary suturing. There were no deaths or complications resulting from the single-stage operation. When a hepatic hydatid cyst is present beneath the diaphragm, the optimal choice is to remove both the liver and lung lesions in a single surgical procedure. We assert that patients with a ruptured intrathoracic hepatic hydatid cyst require medical treatment following surgery, especially if there is dissemination or if complete cyst evacuation is not feasible. We recommend administering albendazole 400 mg twice daily for 3 to 6 months for these patients.

In conclusion, despite the decrease in the frequency of cystic hydatid disease, the rupture of hydatid cysts into the bronchial tree and pleura remains a severe complication that can result in significant morbidity and mortality. Timely evaluation and prompt surgical intervention are essential, particularly before septic complications arise. We advocate for the thoracophrenotomy procedure as a viable and safe surgical option at all three levels. Only long-term percutaneous drainage should be considered for patients in poor general health. Furthermore, medical treatment with albendazole is recommended when dissemination is confirmed. This study is a retrospective, non-randomized analysis with a small patient population at a single center. Nevertheless, long-term studies involving larger cohorts and extended follow-up periods are needed.

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b) All authors read and completed the final version of the manuscript.

c) Manuscript checked by Scribendi for spelling

d) Informed consent was obtained from all patients

Statement of Ethics:

Written informed consent was obtained from all patients; patients attested that they understood their treatment options, potential complications, expected risks, and potential benefits of treatment. The Institutional Ethics Committee approved the study (Reference No: TUTF-GOBAEK 2022/274).

References:

1. Gerazounis M, Athanassiadi K, Metaxos E, Athanassiou M, Kalantzi N. Bronchobiliary fistulae due to echinococcosis. Eur J Cardiothorac Surg 2002;22:306–308.

2. Zaouche A, Haouet K, Jouini M, El Hachaichi A, Dziri C. Management of liver hydatid cysts with a large biliocystic fistula: multicenter retrospective study. Tunisian Surgical Association. World J Surg 2001; 25: 28-39.

3. El Nakeeb A, Salem A, El Sorogy M, Mahdy Y, Ellatif MA, Moneer A, et al. Cystobiliary communication in hepatic hydatid cyst: predictors and outcome. Turk J Gastroenterol. 2017 Mar;28(2):125-130.

4. Virginio VG, Hernandez A, Rott MB, Monteiro KM, Zandonai AF, Nieto A, et al. A set of recombinant antigens from Echinococcus granulosus with potential for use in the immunodiagnosis of human cystic hydatid disease. Clinical & Experimental Immunology. 2003 May 1;132(2):309-15.

5. Karamustafaoglu YA, Tarladacalisir T, Yoruk Y. Abdominothoracic fistulas due to complicated echinococcosis. Thorac Cardiovasc Surg. 2012 Mar;60(2):131-4.

6. Kontoravdis N, Panagiotopoulos N, Lawrence D. The challenging management of hepatopulmonary fistulas. J Thorac Dis 2014;6(9):1336-1339

7. Kim YS, Rhim H, Sung JH, Kim SK, Kim Y, Koh BH, et al. Bronchobiliary fistula after radiofrequency thermal ablation of the hepatic tumor. J Vasc Interv Radiol 2005;16:407-10.

8. Singh B, Moodley J, Sheik-Gafoor MH, Dhooma N, Reddi A. Conservative management of thoracobiliary fistula. Ann Thorac Surg 2002;73:1088-91.

9. Karabulut N, Cakmak V, Kiter G. Confident diagnosis of bronchobiliary fistula using contrast-enhanced magnetic resonance cholangiography. Korean J Radiol 2010;11:493-6.

10. DeOliveira ML, Winter JM, Schafer M, Cunningham SC, Cameron JL, Yeo CY, et al. Assessment of complications after pancreatic surgery: A novel grading system applied to 633 patients undergoing pancreaticoduodenectomy. Ann Surg 2006; 244: 931-9.

11. Sayek M, Iskender, Onat M, Demiral. Diagnosis and treatment of uncomplicated hydatid cyst of the liver. World J Surg 2001; 25:21-7.

12. Yoon DH, Shim JH, Lee WJ, Kim PN, Shin JH, Kim KM. Percutaneous management of a bronchobiliary fistula after radiofrequency ablation in a patient with hepatocellular carcinoma. Korean J Radiol 2009;10:411-5.

13. Liao GQ, Wang H, Zhu GY, Zhu KB, Xin Lv FX, Tai S. Management of acquired bronchobiliary fistula: A systematic literature review of 68 cases published in 30 years. World J Gastroenterol 2011;17:3842-9.

14. Kontoravdis N, Panagiotopoulos N, Lawrence D. The challenging management of hepatopulmonary fistulas. J Thorac Dis 2014;6(9):1336-1339

15. Ferguson TB, Burford TH. Pleurobiliary and bronchobiliary fistulas. Surgical management. Arch Surg 1967;95:380-6.

16. Vagianos CE, Karavias DD, Kassos SK, Vagenas CA, Androulakis JA. Conservative surgery in the treatment of hepatic hydatidosis. Eur J Surg 1995; 161: 415–20.

17. Kabiri EH, El Maslout A, Benosman A. Thoracic rupture of hepatic hydatidosis (123 Cases). Ann Thorac Surg 2001; 72:1883–6.

18. Athanasiadi K, Kalavrouziotis G, Loutsidis A, Bellenis I, Exarchos E. Surgical treatment of echinococcosis by a transthoracic approach: a review of 85 cases. Eur J Cardiothorac Surg 1988; 14: 134–40.

19. Tocchi A, Mazzoni G, Miccini M, Drumo A, Cassini D, Colace L, et al. Treatment of hydatid bronchobiliary fistulas: 30 years of experience. Liver Int. 2007 Mar;27(2):209-14.

20. Gandhi N, Kent T, Kaban JM, Stone M, Teperman S, Simon R. Bronchobiliary fistula after penetrating thoracoabdominal trauma: case report and literature review. J Trauma 2009;67:143-5.