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Case Report

Complex pleural complications in metastatic cholangiocarcinoma: a case report highlighting massive subcutaneous emphysema, bronchopleural fistula and palliative hemostatic radiotherapy

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ABSTRACT

Cholangiocarcinoma (CCA) is an aggressive biliary malignancy with poor prognosis, typically diagnosed at advanced stages. Common metastatic sites include liver, lymph nodes, and peritoneum, while pleural involvement remains rare and poorly described. Such atypical presentations may lead to significant morbidity and complex clinical scenarios requiring invasive management. This study reports a rare case of metastatic CCA with pleural involvement complicated by severe pleuropulmonary manifestations, including infection, bronchopleural fistula, (BPF) and massive subcutaneous emphysema occurring in the setting of malignant pleural disease and persistent pleural air leak requiring one-way valve drainage, as well as life-threatening intrapleural bleeding successfully managed with palliative hemostatic radiotherapy. This case highlights the diagnostic and therapeutic challenges associated with uncommon metastatic patterns in CCA.

Keywords: Cholangiocarcinoma, Pleural metastasis, bronchopleural fistula, Subcutaneous emphysema, Malignant pleural effusion, Hemothorax, Heimlich valve, Palliative radiotherapy

INTRODUCTION

Cholangiocarcinoma (CCA) is a highly aggressive and heterogeneous malignancy that originates from bile duct epithelium. It accounts for 10-15% of all primary liver cancers and 3% of gastrointestinal malignancies.[1] According to its anatomical site of origin, CCA can be classified into intrahepatic (iCCA) and extrahepatic.[1-3]

CCA is characterized by a late-stage presentation and unfavorable prognosis since it is usually asymptomatic in early stages.[1,4] Most known cases are considered sporadic, however, most reported risk factors are related to conditions that cause bile duct chronic inflammation or biliary stasis. Examples include primary sclerosing cholangitis and fibropolycystic liver diseases, biliary stone disease, cirrhosis, hepatitis B and C, hepatic parasitic infections (*O. viverrini* and *C. sinensis),* congenital disorders that cause dilatation of intrahepatic bile ducts such as Caroli disease, inflammatory bowel disease, pancreatitis, alcohol and tobacco abuse, non-alcoholic fatty liver disease, and metabolic disorders including diabetes and obesity.[1,3]

Regarding metastasis, CCA's most common sites include liver, lymph nodes, and peritoneum due to its tendency for locoregional extension and rich periductal lymphatic drainage.[4,5] On the other hand, malignant pleural

involvement is poorly described in literature due to its lower incidence since hematogenous spread is less frequent.[6,7] In this context, pleural effusion often leads to significant morbidity and mortality which requires therapeutic interventions such as tunneled pleural catheters.[8-10] Therefore, we present a complex case of metastatic CCA with rare pleural involvement, in which malignant pleural disease was associated with a cascade of severe pleuropulmonary complications, emphasizing the diagnostic and therapeutic challenges of managing advanced pleural metastasis in CCA.

CASE REPORT

A 62-year-old man with past surgical history of gastric bypass performed 15 years earlier, biliary pancreatitis treated with cholecystectomy 3 years before presentation, and transgastric endoscopic retrograde cholangiopancreatography 2 years prior; was diagnosed in January 2024 with metastatic stage IV CCA. Initial imaging studies revealed multiple hepatic cystic lesions, including one with suspected rupture, right subdiaphragmatic fluid, right pleural effusion, cystic-neoplastic lesions involving the liver and hepatic hilum, probable peritoneal implants, and lymphadenopathy. A liver biopsy performed on January 11, 2024 confirmed the diagnosis. Additionally, serum tumor markers, including CA 19-9 and CEA, were markedly elevated. During the course of his disease, the patient developed recurrent malignant right pleural effusions and multiple pleuropulmonary complications requiring placement of a tunneled pleural catheter and repeated hospitalizations.

In December 2025, the patient was hospitalized for a right-sided empyema associated with the chronic tunneled pleural catheter. Pleural cultures isolated *P. aeruginosa*, sensitive to cefepime, and he completed 4 weeks of inpatient intravenous cefepime with favorable clinical response. During his admission, he underwent three thoracoscopic procedures for empyema drainage and the later finding of a persistent pleural fistula, including right pulmonary decortication on December 9, 2025. His postoperative course was complicated by progressive right-sided subcutaneous emphysema involving the right hemithorax and proximal upper extremity, later extending to the ipsilateral cervical region and hand. On December 21st persistent air leak was documented, with pleural drainage output of 600 mL. Surgical closure of the right BPF was attempted on December 22, with partial reduction of the subcutaneous emphysema. Bronchoscopy on December 26 identified the fistula; occlusion of the right middle pulmonary lobe produced an approximately 100 mL reduction in the air leak, a finding reproduced twice during the procedure. On December 28, he underwent pulmonary segmentectomy, including resection of trapped lung at the costodiaphragmatic angle, release of the lung from the mediastinal pleura, and segmental resection of the anterior segment of the right upper lobe. He was discharged on December 31, 2025 with a Heimlich valve for partial management of the persistent fistula.

The patient was readmitted on January 2, 2026 due to a 24-hour history of increasing swelling of the right hemithorax, extending to the right upper extremity, neck, face, and eyelids. This occurred in the context of known malignant pleural involvement, persistent BPF, and ongoing pleural air leak while the patient was being managed with a Heimlich valve, without a single mechanical cause being definitively established. On admission, he was found to have extensive subcutaneous emphysema and a pleuro-pulmonary fistula with an air leak greater than 500 mL/min. Chest radiographs obtained on January 4 and 5, 2026 showed extensive bilateral subcutaneous emphysema, predominantly on the right side, with partial expansion of right lung and pleural drainage catheter in place (Figure 1 A and B). During this hospitalization, CA 19-9, 25-hydroxyvitamin D (Table 1) and HER2/neu immuno histochemistry from metastatic pleural carcinoma tissue were requested. Bronchoscopy with 7 Fr Swan-Ganz catheter balloon occlusion was performed to localize air leak. All segments and main bronchi of lower, middle, and upper lobes were occluded without evidence of reduction of pleural air leak, despite repeating maneuver twice. Patient subsequently managed with continuous suction pleural drainage, evolved presenting symptomatic improvement and was discharged on Jan 10, 2026 with Thopaz drainage system.

On January 27, 2026 the patient was admitted presenting asthenia, adynamia, general clinical deterioration, progressive dyspnea, and chills. On arrival, he had an irregular tachycardia of approximately 170 beats/min, consistent with atrial fibrillation with rapid ventricular response shown on electrocardiogram (Figure 2 A and B), followed by a subsequent tracing compatible with sinus rhythm after clinical management. Laboratory evaluation showed severe anemia with hemoglobin of 6 g/dL (Table 2), associated with high-volume hemorrhagic pleural drainage of approximately 1000 mL/day. The clinical evaluation resulted in the diagnosis of hypovolemic shock secondary to right hemothorax, complicated by paroxysmal atrial fibrillation. On January 30, infection at the thoracic drainage catheter insertion site was suspected and cultures later isolated *K. pneumoniae*. On February 1, pleural drainage cultures also grew *E. faecalis*, reported as susceptible to tested antimicrobial agents. He was treated with antibiotic therapy, antiarrhythmic management, blood transfusions, and supportive care.

Because of persistent intrapleural bleeding with daily hemoglobin decline, the patient required repeated transfusions, at times up to 3 units of packed red blood cells per day. Tranexamic acid was initiated, and radiation oncology was consulted for palliative hemostatic radiotherapy. On February 2, 2026, he underwent thoracoscopy with pleural cavity lavage and closure of the BPF. Hemostatic radiotherapy was planned

for 10 sessions; after receiving 7 sessions without complications intrapleural bleeding stabilized, with no further hemoglobin decline or transfusion requirement for 2 consecutive days. He was discharged home on February 15, 2026 to continue medical management.

The patient's final admission occurred on March 15, 2026, when he presented with one week of worsening general condition, asthenia, adynamia, and progressive dyspnea, followed 4 days later by fever of 38 °C and significant oxygen desaturation. He initially presented to another hospital, where imaging showed recurrent right pleural effusion and a left pulmonary infiltrate. Broad-spectrum antimicrobial therapy was started, new chronic tunneled pleural catheter was placed in right hemithorax, and diagnostic bronchoscopy was performed. He was then transferred for continued management. On arrival, he was saturating 92% with supplemental oxygen at 6 L/min via nasal cannula, with mild nonproductive cough and diffuse crackles in left hemithorax. B-type natriuretic peptide was greater than 1500 pg/mL. Microbiologic evaluation revealed multiple concomitant infections. Pneumonia PCR panel detected *H. influenzae*, pleural infection was consistent with empyema due to extended-spectrum beta-lactamase-producing *K. pneumoniae*, and patient was also diagnosed with *P. jirovecii* pneumonia. Bronchoalveolar lavage galactomannan positive, suggesting probable concomitant fungal infection. Despite antimicrobial therapy and supplemental oxygen, he developed progressive hypoxemic respiratory failure within 48 hours of admission, requiring sedation and endotracheal intubation.

His clinical course was further complicated by vasopressor-dependent multiorgan failure. Given his advanced metastatic malignancy and critical deterioration, goals of care were discussed with his family. A decision of palliative management was made. Patient died on March 27, 2026. Chronological sequence of major clinical events is summarized in Figure 3.

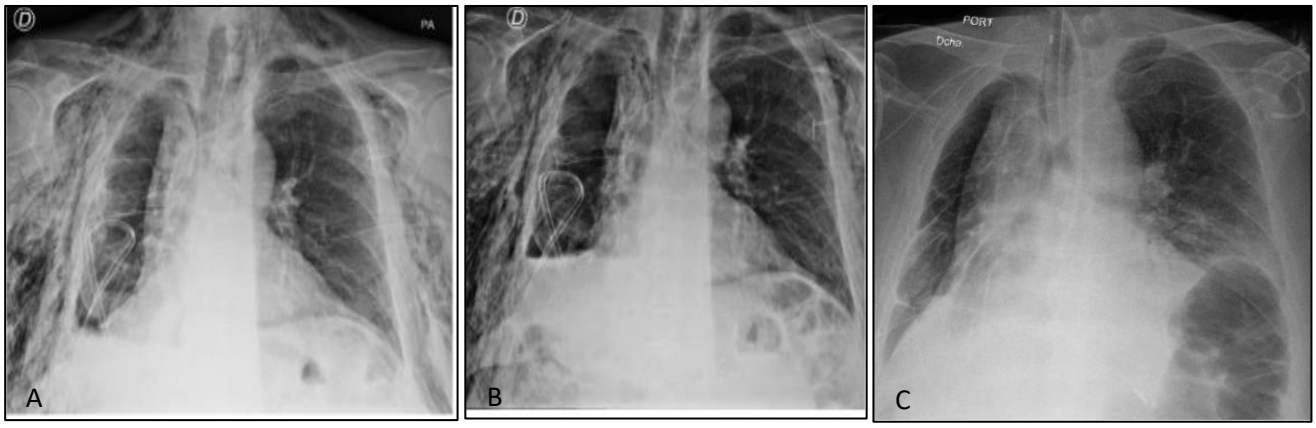

Figure 1 (A-C): Progressive subcutaneous emphysema and persistent pleural disease on serial chest radiographs (A) Jan 4, (B) Jan 5, 2026. C-Persistent pleural disease and reduced right lung volume during final readmission.

*Serial chest radiographs demonstrating progressive subcutaneous emphysema and persistent pleural disease. (A) obtained on January 4, 2026 showing extensive right-sided subcutaneous emphysema with extension into the cervical region and partial expansion of right lung. (B) obtained on January 5, 2026 demonstrating persistent bilateral subcutaneous emphysema, predominantly affecting the right hemithorax, with pleural drainage catheter *in situ.* (C) obtained during the final readmission showing an endotracheal tube in place, persistent right pleural disease with reduced right lung volume, and left basilar/interstitial pulmonary infiltrates.

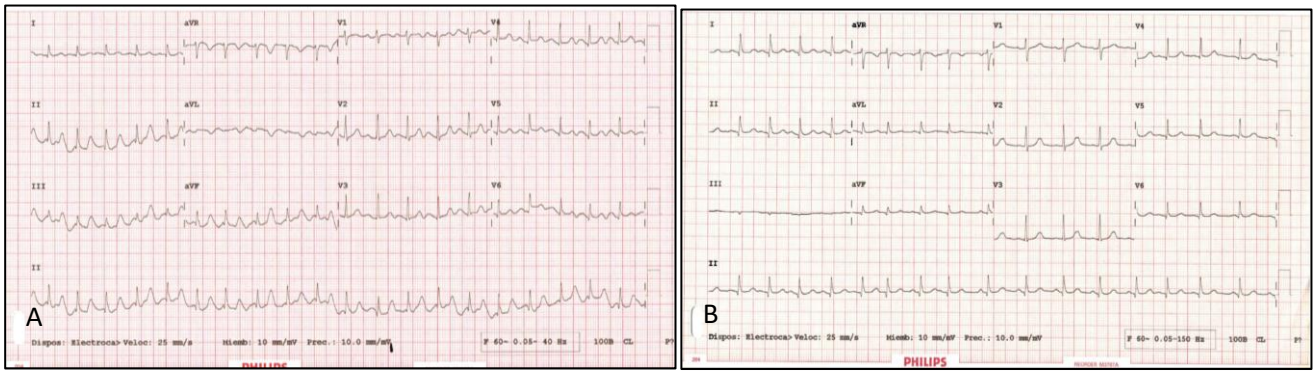

Figure 2 (A and B): A-Atrial fibrillation with rapid ventricular response on initial electrocardiography. B-Restoration of sinus rhythm after clinical management on follow up electrocardiography.

*ECG findings during management for hemorrhagic pleural effusion. (A) ECG demonstrating atrial fibrillation with rapid ventricular response at presentation. (B) Follow-up electrocardiogram showing restoration of sinus rhythm after clinical management.

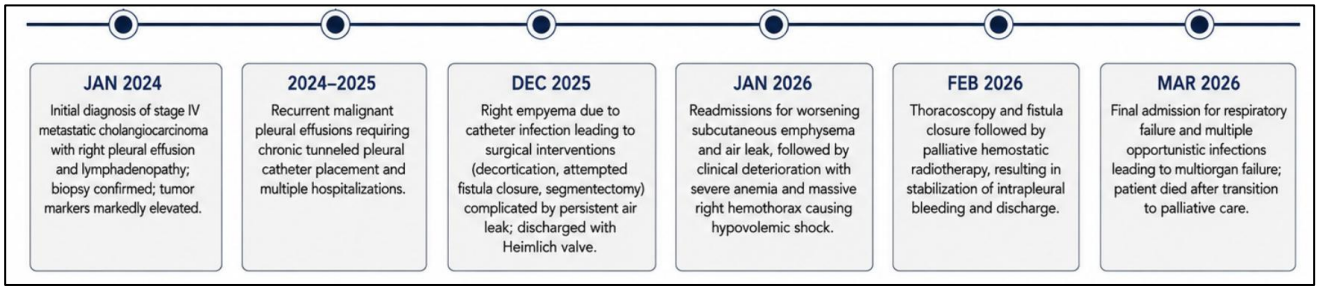

Figure 3: Chronological sequence of major clinical events.

*Chronological timeline of major clinical events during the disease course, including diagnosis of metastatic CCA, development of recurrent malignant pleural effusions, empyema, BPF, massive subcutaneous emphysema, hemorrhagic pleural effusion requiring transfusion support, palliative hemostatic radiotherapy, and final hospitalization complicated by opportunistic infections and multiorgan failure.

Table 1: CA 19-9 and 25-hydroxyvitamin D levels during the first January 2026 readmission.

Results		Units	Reference values	Method
CA 19-9	226.0	U/ml	0.00-39.00	ECL
25-OHD	12.3	ng/ml	30.00-100.00	ECL

Table 2: Blood count findings during the second January 2026 readmission,

Parameters	Results	Units	Reference range
Erythrocytes	1.85	M/µl	4.60-6.00
Hemoglobin	6.0	g/dl	14.0-18.0
Hematocrit	18.1	%	40.0-54.0
MCV	97.8	fL	80.0-94.0
MCH	32.4	pg	26.0-32.0
RDW-CV	16.9	%	11.7-15.5
Leukocytes	5.97	K/µl	4.50-11.50
Neutrophils	54.5	%	50.0-70.0
Lymphocytes	31.2	%	18.0-42.0
Monocytes	12.6	%	2.0-11.0
Eosinophils	1.2	%	1.0-3.0
Platelets	257	K/µl	150-450

DISCUSSION

This case illustrates a rare manifestation of advanced CCA, findings including the occurrence of CCA pleural metastasis complicated by recurrent empyema, persistent BPF with massive subcutaneous emphysema, and transfusion-dependent intrapleural hemorrhage. Additionally, a clinically relevant finding was the temporary control of pleural bleeding following palliative hemostatic radiotherapy. Collectively, these findings illustrate an uncommon and particularly severe manifestation of advanced CCA, underscoring its diagnostic and therapeutic challenges.

CCA is characterized by predominant locoregional spread to the liver, lymph nodes, and peritoneum.[1,4,5] Pleural involvement, however, represents an exceptionally uncommon metastatic manifestation.[6,7] In the population-based SEER analysis by Cheng et al which evaluated site-specific metastases in patients with metastatic intrahepatic CCA, the reported metastatic sites included liver, lung, bone, and brain, without pleural involvement being identified as a common metastatic category.[4] Similarly, Wang et al described the liver, lungs, bone, brain, distant lymph nodes, and peritoneum as the main metastatic sites in extrahepatic bile duct cancer.[5] These population-level findings emphasize the exceptional nature of malignant pleural involvement in CCA and support the unusual clinical presentation observed in this patient.

Published descriptions of pleural involvement from CCA are limited to isolated case reports and conference abstracts. Dziodzio et al described pleural carcinosis during progression of extrahepatic CCA, whereas Ortiz Velez et al reported metastatic CCA presenting with malignant pleural effusion diagnosed by cytology.[6,7] Additionally, evidence from the retrospective series by Yamamoto et al further supported procedure-related tumor seeding as another uncommon mechanism of pleural involvement.[11] The present case differs from the

previously reported because the patient developed a cascade of severe pleural complications, including recurrent empyema, persistent BPF, massive subcutaneous emphysema, and severe intrapleural hemorrhage. Therefore, this case adds to the limited literature by suggesting that malignant pleural involvement from CCA may produce not only recurrent pleural effusions but also multiple overlapping and difficult-to-control thoracic complications.

Indwelling pleural catheters (IPCs) are an established palliative treatment of recurrent malignant pleural effusions because they provide effective symptom relief while allowing outpatient drainage.[8-10] Nevertheless, pleural infection is a recognized complication.[12,13] In the international multicenter retrospective study by Fysh et al involving 1,021 patients with malignant pleural effusion treated with IPCs, pleural infection occurred in 4.9% of patients and most infections were successfully controlled with antibiotic therapy.[12] Similarly, the meta-analysis by Wang et al identified infection as one of the most frequent catheter-related complications, with empyema representing a clinically relevant subset.[13] The clinical course of our patient was considerably more severe than the that reported in previous IPC cohorts. Rather than developing an isolated and antibiotic-responsive catheter infection, he developed recurrent empyema despite prolonged antibiotic therapy and multiple thoracic interventions. Malignant infiltration likely impaired pleural drainage, promoted loculation and chronic inflammation, and reduced tissue healing, creating a favorable environment for persistent infection.[8-10] The comparison with previous IPC cohorts suggests that malignant disruption of the pleural space, may explain this unusually persistent and refractory infectious course.

The development of a BPF represented a major turning point because it resulted in persistent air leakage and extensive subcutaneous emphysema despite multiple surgical and bronchoscopic interventions. BPF is a life-threatening complication associated with prolonged hospitalization, respiratory failure, empyema, and high mortality.[14] Mazzella et al reported an incidence of 0.6%-4% following lobectomy, with mortality rates ranging from 16% to 72%, depending on the clinical setting and timing of diagnosis.[15] Similarly, in a retrospective cohort of 3,180 patients undergoing lobectomy for lung cancer, Ichinose et al identified BPF in 0.44% of patients after lobectomy, and highlighted postoperative inflammation and impaired bronchial healing as important factors associated with its development.[16] Among men undergoing right lower lobectomy, increased risk of BPF was associated with high serum C-reactive protein levels (OR 1.83, 95% CI 1.17-2.87) and previous gastric cancer surgery (OR 5.52, 95% CI 1.06-28.8), whereas bronchial stump coverage was inversely associated with its development (OR 0.06, 95% CI 0.00-0.77).[16] Although these data are largely derived from postoperative lung cancer cohorts, they are clinically relevant because the patient shared pathophysiological conditions known to impair bronchial healing, including chronic pleural infection, persistent inflammation, thoracic interventions, and advanced malignant pleural disease. Unlike the postoperative setting, BPF in this case likely resulted from the combined effects of malignant pleural destruction and recurrent infection rather than from surgical bronchial stump failure alone.

A distinctive feature of this case was refractory intrapleural hemorrhage associated with transfusion-dependent anemia. Although hemorrhagic malignant pleural effusions may occur due to local vasculature disruption, persistent intrapleural bleeding requiring repeated transfusions is rarely described among patients with metastatic CCA.[6-9] In this patient, the hemorrhage was likely multifactorial, resulting from friable tumor-associated vasculature, chronic pleural inflammation, thoracic instrumentation, and impaired tissue integrity. The need for repeated transfusions and tranexamic acid reflects severity beyond the blood-stained effusions commonly encountered in malignant pleural disease. Consistent with previous studies about hemostatic radiotherapy for tumor-related bleeding, our patient experienced stabilization of hemoglobin levels and temporary discontinuation of transfusion requirements following palliative radiotherapy. The retrospective analysis by Guhlich et al reported bleeding control rates of 80-90% with a concomitant reduction in transfusion requirements during radiotherapy, supporting its role as an effective palliative option for clinically significant tumor-related bleeding.[17] However, evidence specifically addressing the use of hemostatic radiotherapy for pleural hemorrhage secondary to metastatic CCA is lacking.

Despite extensive medical, bronchoscopic, surgical, and radiation-based interventions, the patient experienced progressive clinical deterioration, emphasizing the aggressive nature of metastatic CCA and the limited therapeutic options available in advanced stages.[2] This case reinforces the importance of early multidisciplinary management including infectious disease, interventional pulmonology, thoracic surgery, radiation oncology, and palliative care.

This report has limitations inherent to a single case. Causality cannot be definitively established between malignant pleural infiltration, infection, prolonged pleural drainage, repeated thoracic interventions, and the development or persistence of the BPF. Similarly, although subcutaneous emphysema progressed while the patient was managed with a Heimlich valve, the available clinical data do not allow attribution to isolated valve dysfunction. The more likely explanation is multifactorial progression in the setting of malignant pleural disease, persistent BPF, and ongoing pleural air leak. Nevertheless, this case contributes to the limited literature on pleural manifestations of CCA and highlights the significant therapeutic challenges associated with advanced malignant pleural involvement.

CONCLUSION

This case illustrates an unusual and severe pleuropulmonary presentation of metastatic CCA complicated by recurrent empyema, persistent BPF, hemorrhagic pleural effusion, and opportunistic pulmonary infections. CCA with pleural metastasis represents a rare clinical entity, associated with substantial morbidity and risk of life-threatening complications. The present case demonstrates the cascading nature of pleuropulmonary complications in advanced malignancy and highlights the significant diagnostic and therapeutic challenges encountered during management. Additionally, it underscores the potential role of palliative hemostatic radiotherapy as a viable therapeutic option for controlling refractory intrapleural hemorrhage when conventional supportive measures fail. Awareness of these uncommon complications may facilitate earlier recognition, timely multidisciplinary intervention, and improved supportive care strategies. A multidisciplinary approach and early goals-of-care discussions remain essential to guide the patient-centered management according to prognosis and quality of life considerations.

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REFERENCES

1. Banales JM, Marin JJG, Lamarca A, Rodrigues PM, Khan SA, Roberts LR, et al. Cholangiocarcinoma 2020: the next horizon in mechanisms and management. Nat Rev Gastroenterol Hepatol. 2020;17(9):557-88.
2. Vogel A, Bridgewater J, Edeline J, Kelley RK, Klümpen HJ, Malka D, et al. Biliary tract cancer: ESMO Clinical Practice Guideline for diagnosis, treatment and follow-up. Ann Oncol. 2023;34(2):127-40.
3. Zhang H, Yang T, Wu M, Shen F. Intrahepatic cholangiocarcinoma: epidemiology, risk factors, diagnosis and surgical management. Cancer Lett. 2016;379(2):198-205.
4. Cheng R, Du Q, Ye J, Wang B, Chen Y. Prognostic value of site-specific metastases for patients with advanced intrahepatic cholangiocarcinoma: a SEER database analysis. Medicine (Baltimore). 2019;98(49):e18191.
5. Wang X, Yu GY, Chen M, Wei R, Chen J, Wang Z. Pattern of distant metastases in primary extrahepatic bile duct cancer: a SEER-based study. Cancer Med. 2018;7(10):5006-14.
6. Dziodzio T, Jara M, Ritschl PV. Pleural carcinosis of a cholangiocarcinoma. ACG Case Rep J. 2019;6(8):e00146.
7. Ortiz Velez I, Gonzalez Santiago GM, Quiles-Cruz CV, Perez Mitchell MC, Rosado Jimenez M, Font Rivera D, et al. Clinical puzzle unveiled: cholangiocarcinoma in pleural fluid-a rare case report. Am J Respir Crit Care Med. 2024;209(1):A1948.
8. Bibby AC, Dorn P, Psallidas I, Porcel JM, Janssen J, Froudarakis M, et al. ERS/EACTS statement on the management of malignant pleural effusions. Eur Respir J. 2018;52(1):1800349.
9. Gayen S. Malignant pleural effusion: presentation, diagnosis, and management. Am J Med. 2022;135(10):1188-92.
10. Heffner JE, Klein JS. Recent advances in the diagnosis and management of malignant pleural effusions. Mayo Clin Proc. 2008;83(2):235-50.
11. Yamamoto T, Uenishi T, Hirohashi K. Pleural dissemination of cholangiocarcinoma caused by percutaneous transhepatic biliary drainage during the management of resectable cholangiocarcinoma. Surgery. 2019;165(5):912-7.
12. Fysh ETH, Tremblay A, Feller-Kopman D, Mishra EK, Slade M, Garske L, et al. Clinical outcomes of indwelling pleural catheter-related pleural infections: an international multicenter study. Chest. 2013;144(5):1597-602.
13. Wang S, You L, Zheng L. Incidence of complications from indwelling pleural catheter for pleural effusion: a meta-analysis. Clin Respir J. 2022;16(12):780-91.
14. Grotberg JC, Hyzy RC, De Cardenas J, Co IN. Bronchopleural fistula in the mechanically ventilated patient: a concise review. Crit Care Med. 2021;49(2):292-301.
15. Mazzella A, Casiraghi M, Uslenghi C, Orlandi R, Lo Iacono G, Bertolaccini L, et al. Bronchopleural fistula after lobectomy for lung cancer: how to manage this life-threatening complication using both old and innovative solutions. Cancers (Basel). 2024;16(6):1146.
16. Ichinose J, Hashimoto K, Matsuura Y, Nakao M, Okumura S, Mun M. Risk factors for bronchopleural fistula after lobectomy for lung cancer. J Thorac Dis. 2023;15(6):3330-8.
17. Guhlich M, Maag TE, Dröge LH, El Shafie RA, Lang K, Arians N, et al. Hemostatic radiotherapy in clinically significant tumor-related bleeding: excellent palliative results in a retrospective analysis of 77 patients. Radiat Oncol. 2023;18(1):208.

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