CASE REPORT

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Challenges in diagnosing and treating distal common bile duct adenocarcinoma: A case report with literature insights

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Abstract

Background Adenocarcinoma of the distal common bile duct (CBD) is a rare and aggressive malignancy that is often diagnosed at an advanced stage owing to nonspecific symptoms and delayed presentation. This case report details the diagnostic and therapeutic challenges associated with distal CBD adenocarcinoma and highlights the need for an effective multidisciplinary approach.

Case presentation A 54-year-old male with a significant smoking history presented with persistent right upper abdominal pain, dark urine, and scleral jaundice. Imaging studies revealed intrahepatic bile duct dilatation, a mass obstructing the common bile duct, and thickened gallbladder walls. Despite initial antibiotic therapy for suspected cholangitis, the patient underwent endoscopic retrograde cholangiopancreatography (ERCP) and subsequently a surgical procedure. The surgical resection of a common bile duct adenocarcinoma with lymphovascular invasion was successful, with subsequent restoration of bile flow through Roux-en-Y hepaticojejunostomy. Histopathological analysis confirmed tumor characteristics and clear surgical margins. Postoperatively, the patient demonstrated significant clinical improvement with normalized bilirubin levels and received appropriate management for his oncologic condition.

Conclusion This case highlights the diagnostic complexity of distal CBD adenocarcinoma, particularly in patients with delayed symptoms. Multimodal imaging approaches and timely surgical intervention are crucial for effective management of this malignancy. Enhanced awareness of atypical presentations and advancements in targeted therapies holds promise for improving outcomes in such challenging cases.

Keywords Roux-en-Y hepaticojejunostomy, Right upper abdominal pain, Common bile duct adenocarcinoma, Intrahepatic bile ducts, Obstructive jaundice, Lymphovascular invasion

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Background

Adenocarcinoma in the common bile duct exhibits significant heterogeneity, contributing to high mortality and recurrence rates. In Spain, cholangiocarcinomas and adenocarcinomas constitute 57.6% and 42.4% of bile duct tumors, respectively, with perihilar cholangiocarcinomas being the most common (40%) [1]. Most cases (63.6%) are unresectable at diagnosis, emphasizing the need for early detection. Ampullary carcinoma, a rare subtype, shows a higher incidence in males and non-Hispanic Whites, with worse prognosis associated with age and advanced stage at diagnosis. African American and Hispanic populations face higher mortality risks [2, 3]. In Korea, the incidence of distal common bile duct (CBD) cancer among periampullary cancers is higher compared to other countries, with limited data indicating late-stage diagnosis and poor prognosis [3, 4]. Pre-existing conditions such as chronic hepatitis, hepatic steatosis, and cholelithiasis are common risk factors [1, 5].

Patients frequently present with jaundice and elevated CA 19-9 levels, indicating advanced disease and poor prognosis. A Spanish study showed nearly 46% of cases diagnosed at stage IV, underscoring late diagnosis as a common issue [1]. Socioeconomic factors and healthcare access, such as Medicare coverage, also impact diagnosis and treatment accessibility [5]. Imaging modalities are essential in differentiating benign from malignant obstructions, with magnetic resonance cholangiopancreatography (MRCP) and endoscopic ultrasonography (EUS) providing high-resolution soft tissue imaging. MRCP is effective for bile duct anatomy, while EUS, particularly with FNA, enhances cellular analysis accuracy [5]. CT is valuable for staging and identifying metastases, although it's less sensitive in early malignancies. Ultrasound is widely available and often used initially due to its accessibility and cost-effectiveness; however, follow-up imaging with more advanced modalities such as MRCP or CT is frequently necessary to obtain detailed anatomical information and to confirm the diagnosis.

Combining these approaches optimizes diagnostic accuracy and reduces invasive biopsy needs, though his-tological confirmation remains essential [1].

Surgical resection remains the primary curative approach, with pancreaticoduodenectomy (Whipple procedure) aiming for R0 resection in resectable distal bile duct tumors, significantly improving survival prospects. Extended hepatectomy is preferred for proximal involvement to ensure clear margins [6]. For unresectable tumors, palliative options, such as biliary stenting, managing symptoms and improving the quality of life [7]. Adjuvant therapies, including gemcitabine and cisplatin, along with emerging targeted therapies (e.g., FGFR and IDH inhibitors), show promise for tumors with specific mutations [8]. Immunotherapy may benefit patients with PD-1 or PD-L1 expression.

Documenting rare presentations of common bile duct adenocarcinoma is essential for enhancing diagnostic accuracy and therapeutic planning. Expanding clinical understanding through such cases aids in recognizing atypical symptoms or imaging findings, potentially leading to the earlier identification of malignancies. This broader knowledge base refines prognostic models and guides research into novel biomarkers or genetic factors, ultimately promoting personalized treatment approaches for complex disease profiles [9].

This case report describes a rare presentation of adenocarcinoma in the distal common bile duct with a delayed diagnosis, highlighting the clinical and diagnostic challenges associated with atypical symptom progression. By detailing the comprehensive diagnostic approach, surgical intervention, and postoperative management, we sought to provide insights that can improve early detection and tailored therapeutic strategies for similar complex cases.

Case presentation

A 54-year-old married male with a 36-pack-year smoking history presented with complaints of persistent right upper abdominal pain that was unrelieved by analgesics, accompanied by dark urine and scleral jaundice. These symptoms persisted for three months, with a reported weight loss of 10 kg over the last two months. The patient did not seek medical attention until the symptoms worsened. On initial assessment, his vital signs were stable, and his medical history included hypertension, for which he was on antihypertensive medication. The patient had no significant surgical history.

Abdominal ultrasonography revealed intrahepatic bile duct dilatation and a distended common bile duct (CBD) measuring 20 mm with an intraluminal heterogeneous mass near its distal portion. The gallbladder was filled with a biliary sludge. The patient was admitted to the gastroenterology department for further treatment. Initial blood tests showed elevated total bilirubin (17.8 mg/dL) and direct bilirubin (9.3 mg/dL) levels. Given the suspicion of cholangitis, an empirical antibiotic therapy was initiated. However, the patient did not show any significant improvement.

EUS was performed and revealed thickening of the bile duct.

The patient subsequently underwent ERCP, which revealed a normal duodenum. A suspected stone or mass, approximately 5 mm in size, was observed in the CBD at the mid-lower third, obstructing flow through the midupper third and preventing the contrast material from passing toward the liver. Multislice computed tomography (MSCT) revealed dilation of the biliary tract, with CBD measuring 17 mm, common hepatic duct (CHD) measuring 14 mm, and intrahepatic ductal dilation (see Fig. 1). Stones, masses, and suspicious lesions were not observed within these structures.

MRCP revealed significant stenosis at the confluence of the right and left hepatic ducts, with marked intrahepatic ductal dilatation. Gallbladder wall thickening (12 mm), along with biliary sludge and small gallstones, were also noted. Percutaneous transhepatic biliary drainage (PTBD) was performed to lower the bilirubin levels and stabilize the patient for surgery. Intraoperatively, a mass was discovered in the CBD, which was in contact with the portal vein and hepatic artery. The tumor was resected with a 1 cm safety margin superiorly and a 2 cm margin inferiorly (**see** Fig. 2), and cholecystectomy was performed.

The tissue specimens were sent for histopathological evaluation. Roux-en-Y hepaticojejunostomy was performed to restore bile flow and to connect the remaining portion of the CBD to the jejunal loop. The surgery concluded with jejunostomy creation and wound closure.

The histopathological examination of the samples revealed significant findings. An adenocarcinoma in the common bile duct was discovered, extending 5 mm into the duct wall, with lymphovascular invasion (LVI). The absence of any excised lymph nodes made it impossible to perform lymph node staging. The presence of LVI was confirmed intraoperatively by observing tumor emboli within the lymphatic and vascular channels adjacent to the resected mass. The microscopic examination revealed extensive chronic inflammatory infiltrates permeating all layers of the duct wall, accompanied by notable ulceration. Additionally, there was significant tumoral proliferation characterized by the presence of tubules, papillae, and glands. These structures were lined with large, polygonal, or stratified cells that exhibited hyperchromatic nuclei. The surgical margins were confirmed to be free of the tumor.

After surgery, the patient demonstrated significant clinical improvement with normalization of the bilirubin levels. Postoperative management included prophylactic anticoagulation due to the patient's oncologic status, limited movement to reduce the risk of thromboembolism, analgesics, and gastric protectants. The plan also included improving the patient's overall health and nutritional status, follow-up with a gastroenterologist, and referral to an oncology hospital for a multidisciplinary approach.

It showed mild dilation of the CBD (17 mm), CHD (14 mm), and pancreatic duct (2.2 mm), with no visible stones or suspicious masses. The liver appeared homogenous with mild fatty infiltration and the gallbladder contained biliary sludge. No free fluid or suspicious lymphadenopathy were detected in the abdomen or pelvis.

The cholecystectomy specimen revealed a gallbladder with dimensions of $9.8 \times 4.5 \times 1.6$ cm and a surgical rupture measuring 1 cm. Upon opening, eroded mucosa with hemorrhage was noted, with wall thickness ranging from 0.2 to 1 cm. No calculi were found. Additionally, the CBD was examined, measuring 2.7×1.2 cm, and a 4×1.6 cm cyst was identified. Upon opening, a septate cyst with wall thickness of 0.3 cm was observed.

Discussion

Delayed diagnosis is a common challenge in cancers such as distal CBD adenocarcinoma and has been shown to significantly worsen prognosis by allowing progression to advanced stages before treatment initiation. This pattern is consistent with that of other gastrointestinal cancers,

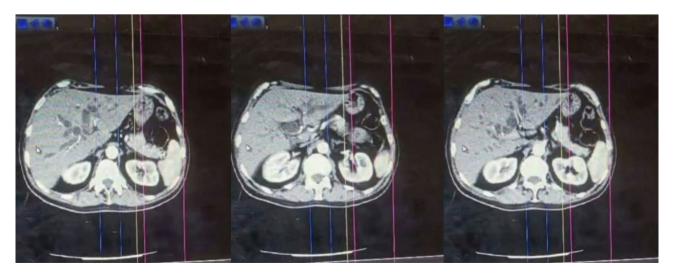


Fig. 1 MSCT scan of the abdomen and pelvis

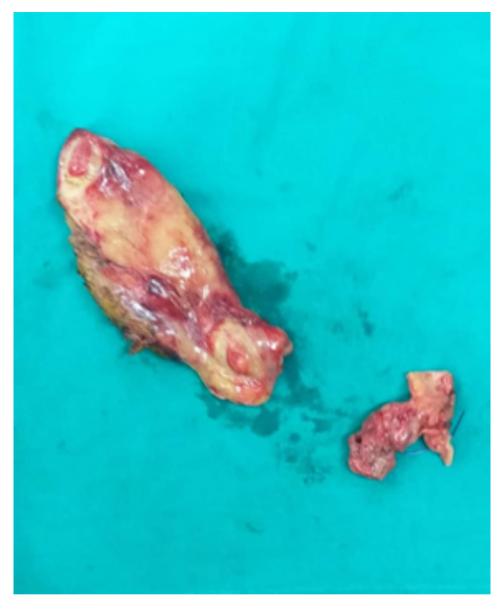


Fig. 2 Cholecystectomy and common bile duct

such as gastric adenocarcinoma, where delays in diagnosis and treatment initiation are linked to reduced survival rates [10]. Similarly, colorectal and pancreatic cancers also demonstrate the detrimental effects of delayed diagnosis, which often leads to advanced tumor stages at the time of detection [11, 12].

The upper abdominal pain is the main presentation for most cases recorded in medical literature [13]. In our case, the patient's presentation with persistent right upper abdominal pain, jaundice, and significant weight loss over a three-month period before seeking medical care reflected a delayed diagnosis trajectory. This delay in recognizing cancer likely contributed to the advanced stage of the disease upon diagnosis, aligning with findings from similar studies on delayed presentations of bile duct cancers. Early detection and intervention, as seen in the aforementioned studies, are crucial for improving survival outcomes, underscoring the need for timely diagnostic and therapeutic efforts.

Accurate diagnosis of distal CBD adenocarcinoma necessitates the use of precise imaging techniques, which are often challenging owing to the limitations of common diagnostic tools. It is difficult to detect small tumors or distal bile duct lesions using ultrasound, which is commonly used as an initial imaging method, due to limited resolution and interference from bowel gas [14]. In our case, abdominal ultrasonography identified intrahepatic bile duct dilation and a distended CBD with an intraluminal heterogeneous mass. ERCP has been integral in diagnosing pancreatic diseases and obtaining detailed

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imaging of the pancreatic duct. With advancements in technology, it has evolved into a vital tool for therapeutic stenting and various treatment-oriented applications, including collecting pure pancreatic juice and performing intraductal ultrasonography [15]. Similarly, while ERCP can offer direct visualization and tissue sampling, it carries risks, such as pancreatitis, and may not detect small mucosal lesions [16, 17]. Previous studies have shown that CT imaging has demonstrated high accuracy in distinguishing between benign and malignant periampullary tumors, underscoring its critical role in diagnosis and treatment planning. Additionally, CT scans are valuable in identifying biliary obstruction and evaluating the resectability of lesions, emphasizing its importance in comprehensive vascular assessment [18]. The use of MSCT in our case provided important anatomical insights showing ductal dilation but no visible masses, aligning with the limitations noted in the literature [16, 17]. MRCP, used in our patient to evaluate biliary tree involvement, is highly useful for detecting stenosis, but may lack the specificity to differentiate between benign and malignant strictures [16, 17]. The combined use of multiple imaging modalities, as observed in our patient's diagnostic workup, can improve diagnostic accuracy and help better evaluate complex cases of bile duct adenocarcinoma.

Percutaneous transhepatic biliary drainage (PTBD) and stenting are effective treatments for malignant obstructive jaundice with minimal complications, making them ideal for elderly patients such as in our patient [19]. Surgical resection, especially through pancreaticoduodenectomy (Whipple procedure), remains the mainstay of treatment for resectable distal CBD adenocarcinomas. However, the use of Roux-en-Y hepaticojejunostomy (RYHJ) to restore bile flow after resection has gained prominence in several studies. One study highlighted the use of RYHJ in patients treated for perihilar cholangiocarcinoma, showing its potential to reduce surgical complexity and prevent adhesions [20]. Similarly, a robotic approach to RYHJ in bile duct cancer patients has been linked to improved postoperative outcomes, including a lower risk of anastomotic strictures and cancer recurrence [21]. In our case, the patient underwent a successful RYHJ following resection of the distal CBD tumor, which contributed to bile flow restoration and significant postoperative improvement. These studies underscore the potential benefits of RYHJ, particularly in complex biliary reconstructions and advanced cancers, as observed in our patient's favorable postoperative outcomes.

Lymphovascular invasion (LVI) is a critical prognostic factor in bile duct adenocarcinoma that influences both disease-free survival (DFS) and overall survival (OS). Studies have shown that LVI is associated with poorer outcomes in distal cholangiocarcinoma, with patients exhibiting higher LVI levels and significantly reduced survival rates [22]. In our case, histopathological examination revealed lymphovascular invasion within the tumor, which was indicative of a more aggressive disease course. This aligns with findings from pancreatic adenocarcinoma and other gastrointestinal cancers, in which LVI serves as a strong indicator of poor prognosis [23]. The presence of LVI in our case underscores the need for aggressive postoperative treatment strategies including adjuvant therapies to improve survival and reduce recurrence.

Postoperative outcomes for patients with distal CBD adenocarcinoma undergoing pancreaticoduodenectomy have shown improvements in recent years, particularly in terms of reduced blood loss, shorter operation times, and lower infection rates [4]. However, complications, such as postoperative pancreatic fistulas, remain challenging. Our case reflects this reality, as the patient demonstrated significant clinical improvement post-surgery, including normalization of bilirubin levels; however, such outcomes require careful postoperative management. Studies have shown that factors such as advanced tumor stage and high preoperative CA19-9 levels continue to affect survival, highlighting the importance of early detection and comprehensive perioperative care [4]. As observed in our patient, careful management of biliary drainage and minimization of postoperative complications are crucial for achieving a favorable outcome.

Early detection is paramount for improving survival rates and reducing recurrence in bile duct cancers. Advancements in biomarkers, such as circulating tumor DNA and microRNA, show promise for early stage diagnosis, enabling earlier interventions that may result in curative outcomes [24]. In our case, the patient's prolonged symptoms and delayed diagnosis emphasized the need for better early screening methods. Technologies such as endoscopic ultrasound (EUS) have been shown to reduce recurrence rates by enabling earlier detection and targeted intervention [25]. Early detection, especially in high-risk populations, significantly improves the chances of curative treatment, as demonstrated by studies on cholangiocarcinoma [26]. For our patient, more timely screening could potentially lead to earlier intervention, improving long-term outcomes.

Emerging therapies for the treatment of distal CBD adenocarcinoma, particularly in advanced stages, are beginning to show promising results. Targeted therapies, such as PARP inhibitors for BRCA-mutated tumors and KRAS inhibitors for KRAS-mutant cancers, are being explored to extend progression-free survival [27]. In addition, photodynamic therapy (PDT) combined with RNA-based medicines is emerging as a novel strategy to treat fibrotic tumors such as pancreatic and bile duct cancers [28]. Biomarker-driven therapies are also advancing, with the use of immune checkpoint inhibitors in mismatch repair-deficient tumors and NTRK inhibitors in NTRK fusion-positive tumors; these have seen regulatory approval in specific subtypes, marking steps forward in personalized medicine for these cancers [29]. These advancements highlight the potential of personalized medicine to improve outcomes in patients with advanced CBD adenocarcinomas. While these treatments are still in the early stages, they offer optimism for more effective management of distal CBD adenocarcinomas in the future.

Conclusion

This case highlights the complexities of diagnosing and managing distal common bile duct CBD adenocarcinoma, a rare and aggressive cancer often identified only in advanced stages owing to vague symptomatology and challenging imaging findings. The successful surgical intervention and postoperative improvement in our patient underscores the value of a multidisciplinary approach that incorporates advanced imaging and targeted surgical techniques to achieve favorable outcomes. Early detection remains crucial, as delayed diagnosis typically correlates with poor prognoses. Future advancements in diagnostic tools, including biomarkers and molecular imaging, along with emerging targeted therapies, offer hope for earlier detection and personalized treatment pathways for patients with distal CBD adenocarcinoma.

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Author contributions

S.A., H.H., Y.A., M.A., H.A., M.A.N., T.A.A., and B.A. wrote the main manuscript text. Dr. Mohammad Shafa'a and Dr. Muhammad Fadi Alkurdi conceived and supervised the conduct of the study. All authors critically reviewed and revised the manuscript.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

Ehical clearance was obtained from the ethical committee of Kalamoon University (Approval Number: 350) and consent was obtained from our patient to prepare the case for case report.

Consent for publication

Written informed consent was obtained for the publication of this case report.

Competing interests

The authors declare no competing interests.

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