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Case Report

A Rare Complication of Biliary Pancreatitis Forming a Radiological Mass Mimicking a Klatskin's Tumor

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Abstract: A Klatskin tumor, a type of cholangiocarcinoma that develops where the right and left hepatic bile ducts join to form the common bile duct. These tumors, first identified by Altemeier et al. in 1957 and later elaborated upon by Klatskin in 1965, typically occur within 2 cm of the hilar confluence and account for 50–70% of all cholangiocarcinomas. It is important to differentiate Klatskin tumours from various benign conditions and other malignant lesions that can mimic both the clinical presentation and radiological appearance, often termed Klatskin-like lesions. There have been multiple reports of hilar strictures initially diagnosed as cholangiocarcinoma and it has been found that 5-15% of these cases ultimately exhibit benign lesions upon final histopathological examination. To our knowledge there is no case of a complication of biliary pancreatitis leading to the formation of a radiological lesion mimicking a Klatskin's like tumor, being reported in the literature. Therefore, this case report represents the first documented instance of this intriguing occurrence.

Keywords: Biliary Pancreatitis; Klatskins Like Tumor; Cholangiocarcinoma.

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1. Introduction

A Klatskin tumor, a type of cholangiocarcinoma that develops where the right and left hepatic bile ducts join to form the common bile duct. These tumors, first identified by Altemeier et al. in 1957 and later elaborated upon by Klatskin in 1965, typically occur within 2 cm of the hilar confluence and account for 50–70% of all cholangiocarcinomas [1]. It is important to differentiate Klatskin tumours from various benign conditions and other malignant lesions that can mimic both the clinical presentation and radiological appearance, often termed Klatskin-like lesions [1, 2].

There have been multiple reports of hilar strictures initially diagnosed as cholangiocarcinoma and it has been found that 5-15% of these cases ultimately exhibit benign lesions upon final histopathological examination [3]. To our knowledge there is no case of a complication of biliary pancreatitis leading to the formation of a radiological lesion mimicking a Klatskin's like tumor, being reported in the literature. Therefore, this case report represents the first documented instance of this intriguing occurrence.

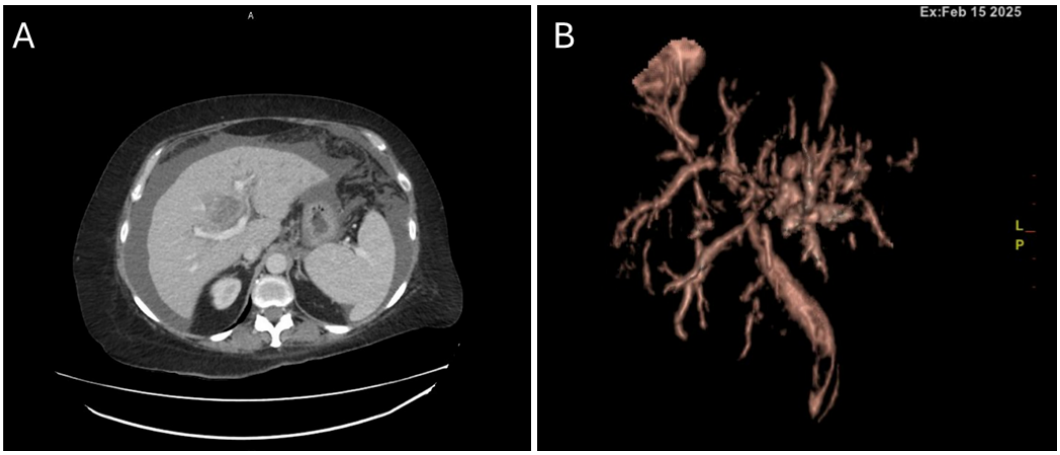
2. Case Report

A 60-year-old female patient with a history of biliary pancreatitis, status post ERCP and stenting, and biliary sphincterotomy complicated by pancreaticoduodenal fistula s/p cholecystectomy, presented to the ED due to rising billirubin levels detected in her OP blood work. On presentation patient endorsed significant weight loss of 80lbs associated with malaise and fatigue. She denied fevers, abdominal pain, vomiting, diarrhea, chest

pain, shortness of breath/cough. She appeared icteric. Abdominal examination was not remarkable. LFTs were deranged AST/ALT-206/64, ALP-649, Total Billirubin 9.8 and Direct Billirubin 7.6 suggestive of a cholestatic jaundice. Tumor markers CA 19-9-102 which decreased from 290 a month prior, CEA-1.

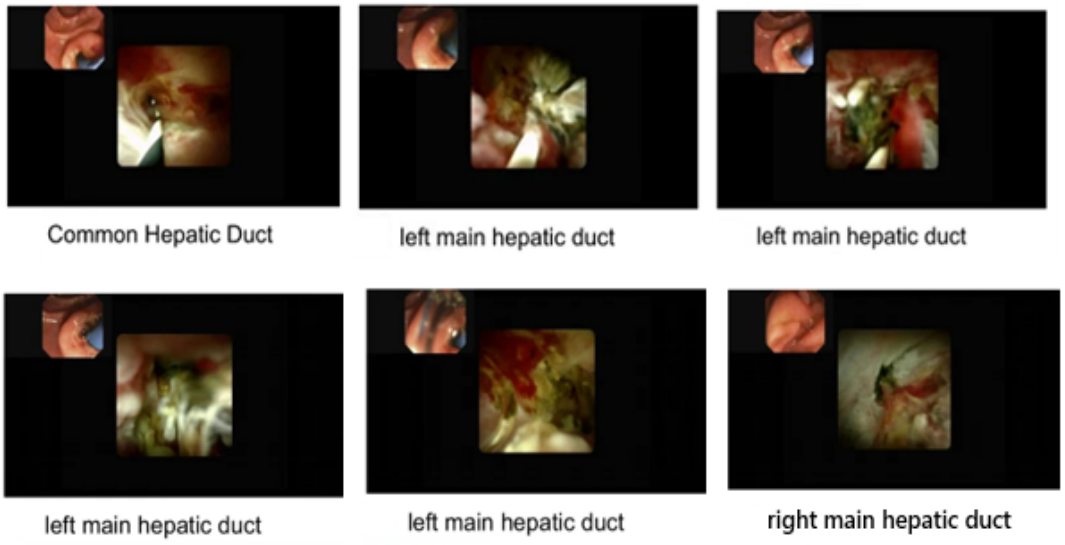
In the ED, a CT abdomen revealed known pancreatitis changes along with a mass in the porta hepatis 59 x 54 x 40 mm, concerning Klatskin's tumor. Left hepatic ductal dilation and abdominal ascites were noted (Figure 1A). MRCP performed on the same day showed a large lobular, hypoechoic but heterogenous enhancing mass measuring 58 x 39 x 78 mm, causing mild-to-moderate left and mild right intrahepatic biliary dilation with a non-dilated lower CBD (Figure 1B). There was also mild periportal lymphadenopathy and mild to moderate diffuse ascites which appeared to be malignant ascites given the remainder of the findings. This raised a strong concern for Klatskin's tumor.

Figure 1. CT abdomen and MRCP showing a mass in the porta hepatis.



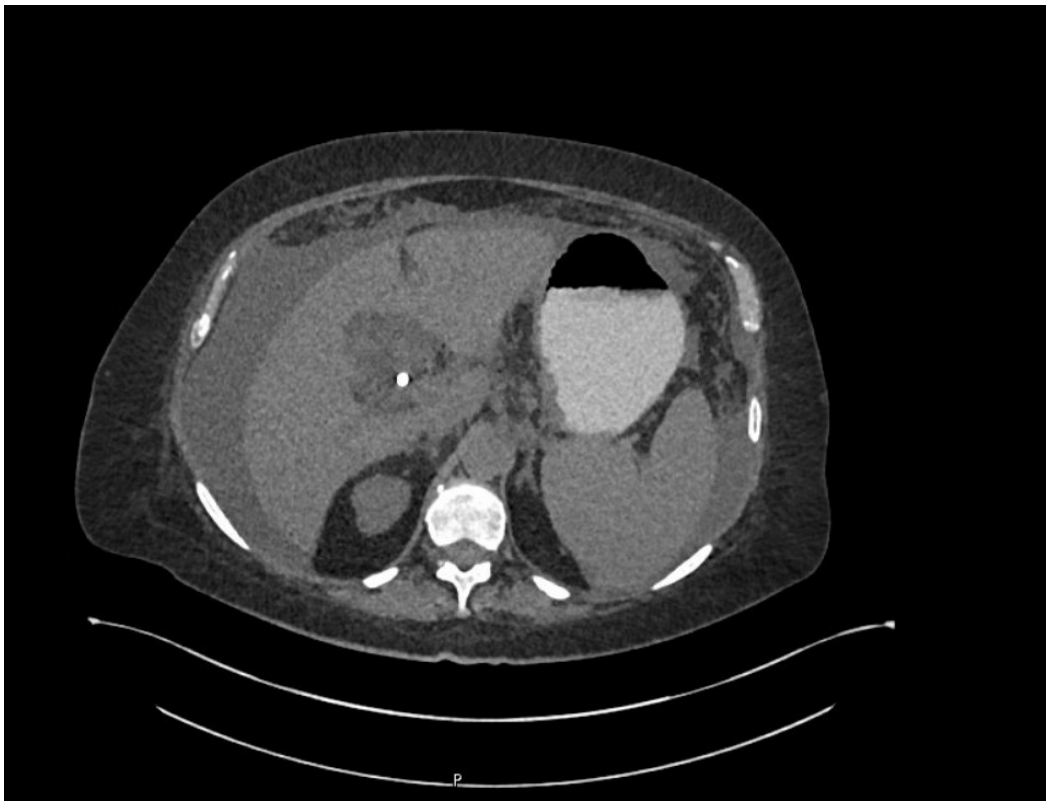
An ERCP/EUS was also conducted, but the mass could not be biopsied due to its location. A percutaneous biopsy was deferred due to concerns of potential seeking if the mass was indeed malignant. A subsequent sphincterotomy was performed with balloon extraction of choledocholithiasis and stenting of the CBD. A repeat ERCP with cholangioscopy with biopsy and cytology brushing was performed in the left and right hepatic ducts which were subsequently negative for malignancy (Figure 2).

Figure 2. ERCP performed during which brush biopsies were collected.



A repeat CT A/P a month following discharge re-demonstrated the lesion in the porta hepatis which was now markedly enlarged measuring approximately 52 x 51x 68 mm with a clear increase in its craniocaudal extent and a more lobulated, ill-defined contour. There was no new vascular encasement or worsening intrahepatic biliary ductal dilation. The lesion did not demonstrate significant enhancement on arterial sequences and heterogeneous internal enhancement on venous phases. There was no rim enhancement to suggest abscess. This rapid change in size and heterogeneous contour favored an infectious/inflammatory process rather than a neoplastic process (Figure 3).

Figure 3. CT abdomen a month later showing increased size of the porta hepatis mass.



3. Discussion and Conclusion

This case report describes a rare instance of a complication of biliary pancreatitis leading to the formation of a radiological lesion mimicking a Klatskin's like tumor, which is the first such case reported in the literature. Kumar et al. in their study document the diverse etiologies of klatskin like lesions, including inflammatory (e.g., IgG4-related cholangiopathy, eosinophilic cholangiopathy), infectious (e.g., AIDS cholangiopathy, biliary tuberculosis), vascular conditions (e.g., portal hypertensive biliopathy), toxic (e.g., post-chemotherapy effects), traumatic, and neoplastic causes (both malignant, such as hepatocellular carcinoma, and benign, such as neurilemmoma). Additionally, miscellaneous conditions like Erdheim-Chester disease and idiopathic cases contribute to the spectrum of Klatskin-mimicking lesions [1].

In IgG4 related cholangiopathy there are elevated IgG4 levels with biliary stricturing often accompanied by autoimmune pancreatitis whereas in eosinophilic cholangiopathy, there is associated eosinophilia, both of which were absent in our patient [4]. To our knowledge, a localized porta hepatis inflammatory tumor arising as a complication of biliary pancreatitis has not been previously described.

Both malignant and benign biliary structures present with similar clinical symptoms, including jaundice, pruritus, dark urine, pale stools, abdominal discomfort, and fatigue.

Additionally, bilirubin levels and serum tumor markers such as carbohydrate antigen 19-9 (CA19-9), interleukin-6 (IL-6), and neutrophil gelatinase-associated lipocalin (NGAL) lack sufficient diagnostic accuracy to reliably distinguish between malignant and benign strictures [5]. The elevated CA19-9 in our patient was likely a false positive related to her history of acute recurrent pancreatitis and cholestatic jaundice rather than a neoplastic etiology. Tsalis et al. in their study note that some lesions that mimic Klatskin tumors, such as tuberculosis, sarcoidosis, lymphoma, and metastasis, may also present with lymphadenopathy, as in the present case and lead to a potential misdiagnosis [6].

In the present case, initial radiological imaging including CT scan and MRCP were suggestive of Klatskin's tumor. The diagnostic challenge in this case stemmed from the radiological appearance of the intrahepatic mass, which closely resembled malignancy, but histopathological examination and repeat radiological tests revealed the lesion was likely benign, stemming from an infectious/inflammatory etiology due to biliary pancreatitis. Our hypothesis is that recurrent biliary pancreatitis combined with post ERCP sphincterotomy, stenting and pancreaticoduodenal fistula created a permissive route for enzyme rich pancreatic fluid to travel along the hepatoduodenal ligament to the porta hepatis. This likely resulted in an intense inflammatory process, necrosis and subsequent organization into an inflammatory pseudo tumor in the hilar area. The location of this mass led to a radiological appearance indistinguishable from a Klatskin's tumor.

An important limitation of this case is the absence of a core biopsy of the hilar mass which was deferred due to concerns of tumor seeding. As a result, our diagnosis of an inflammatory pseudo tumor related to biliary pancreatitis remains presumptive and is based on the negative intraductal histology combined with clinical and radiological evolution. This highlights the importance of considering a broad differential diagnosis when encountering masses near the hepatic hilum to avoid misdiagnosis, undue emotional distress for patients and unnecessary surgical intervention. Although, this can be achieved through thorough clinical assessment and multimodal non-invasive radiological diagnostic methods, invasive histopathological modalities remain the mainstay for conclusive diagnosis in many cases.

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Research Ethics Committee Approval: The patient provided written informed consent to participate in the study, which was conducted in accordance with the ethical principles of the Declaration of Helsinki.

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Conflicts of Interest: All other authors declare no conflicts of interest.

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