

“Travel Jaundice” Caused by Pathological Common Bile Duct Angulation: A Case Report

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Abstract

Background: Physiologically, the common bile duct (CBD) turns to the right as it descends towards the duodenum. The degree of angulation in this turn is dependent on the position of the patient, liver mobility and points that stabilize the CBD, such as postoperative adhesions. In extreme cases, the sharpness of this angulation can by induce jaundice and recurrent choledocholithiasis. In this report, the authors describe a woman for whom pathological angulation occurs only in a sitting position. In view of periodic appearance of symptoms to establish the correct diagnosis it is difficult, therefore we proposed a method of enabling confirmation of the existence of this unusual pathology. In the available publications we not found a similar description of the disease and methods for its diagnosis.

Case presentation: The paper presents the a 55-year-old woman with recurrent pain in the right upper quadrant with comorbid jaundice most often after a long journey by car. In the past patient underwent classical cholecystectomy with revision of the CBD and endoscopic sphincterotomy. Despite this treatment, symptoms continued to recur. On that basis, we decided examine her biliary tree in a sitting position using for this purpose nasobiliary tube (NBT). The image acquired in this way revealed the presence of a pathological CBD angulation, which explained the symptoms.

Conclusion: Bending to the bile duct is rare and difficult to diagnose cause transient jaundice. However, in some cases where the clinical picture points to the cause, cholangiography using the NBT can be the basis to determine the correct diagnosis.

Keywords: Common bile duct; Travel jaundice; Comorbid jaundice

Abbreviations: CBD –Common bile duct; RUQ -right upper quadrant ; ERCP -endoscopic retrograde cholangiopancreatography ; NBT -nasobiliary tube. SOD- Sphincter of Oddi dysfunction ; APBDU -anomalous pancreaticobiliary duct union. CTC-computed tomography cholangiography. MRCP-magnetic resonance cholangiopancreatography.

Introduction

Physiologically, the common bile duct (CBD) curves to the right as it descend toward the duodenum; this curvature can cause intermittent problems with respect to the outflow of bile [1]. The degree of angulation in this curve is dependent on liver mobility and the existence of points of CBD stabilization, such as postoperative adhesions or tumors. In extreme cases, the sharpness of this angulation can lead to periodic jaundice and recurrent choledocholithiasis. Keizman first described pathological angulation of the CBD in a study in which the angle between the portions of the CBD located 1 cm below the bifurcation of the bile ducts and the portion of the CBD 1 cm above the papilla of Vater was measured with the patient in the prone position [2]. He defined pathological angulation as the presence of an angle sharper than 145 degrees [1,3]. A slightly different definition of this pathology was offered by Kim, who measured the angle between the medial and distal CBD segments and concluded that an angle sharper than 135 degrees was pathological [3]. In this study, the authors present an extremely rare case involving a patient in whom pathological CBD angulation causes temporary obstructive jaundice only after a prolonged car trip in a sitting position. Issues regarding the confirmation of this unusual diagnosis were also analyzed.

Case Report

A 55-year-old woman was admitted to the surgical ward because of recurrent pain in the right upper quadrant (RUQ) with comorbid jaundice. The patient's anamnesis revealed that she had suffered from similar symptoms on several occasions after a prolonged journey by car; in these cases, symptoms generally persisted for 2-3 days

and spontaneously resolved within the subsequent 4-5 days. An examination of similar cases from the previous 10 years indicated that patients were treated using the classical approach of a cholecystectomy with biliary duct revision due to choledocholithiasis and T-tube drainage of the CBD. For the current patient, endoscopic retrograde cholangiopancreatography (ERCP) had been performed two years earlier because of the aforementioned symptoms. During this procedure, choledocholithiasis was excluded, and an endoscopic sphincterotomy was performed because the clinical picture suggested Sphincter of Oddi dysfunction (SOD). Despite this treatment, symptoms continued to recur, with each episode of symptoms occurring after a prolonged car trip. Physical examination at admission revealed a slight yellowing of the eyes and tenderness in the RUQ. Biochemical examinations indicated that the patient had elevated levels of bilirubin (2.0/1.8 mg/dl, normal 1.2 mg/dl) but low levels of serum amylase (45/36 U/L, normal 80 U/L), urinary ERCP had been performed two years earlier because of the aforementioned symptoms. During this procedure, choledocholithiasis was excluded, and an endoscopic sphincterotomy was performed because the clinical picture suggested Sphincter segment of the CBD that was distended to 10 mm without biliary stones. The pancreas was normal in size but contained numerous small calcifications indicative of chronic inflammatory changes. No pathological findings were observed in other examined organs. The patient was re-qualified for ERCP, which indicated that the CBD had a diameter of approximately

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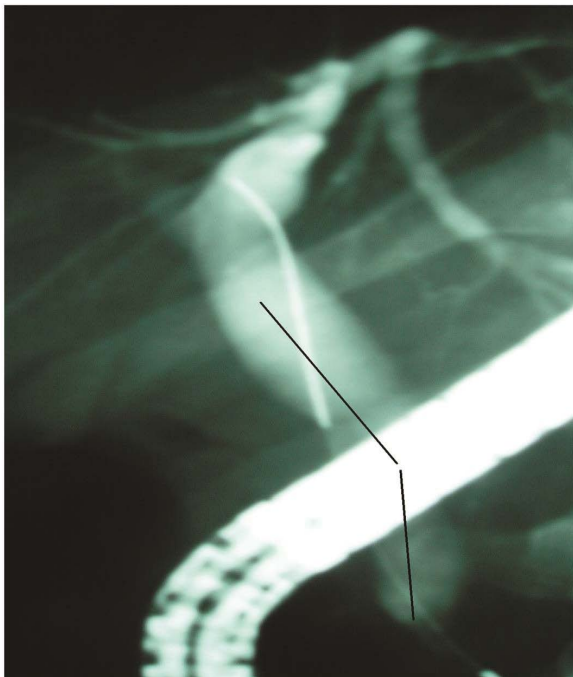


Figure 1: The CBD had a diameter of approximately 11 mm, with appropriate contrast in outflow to the duodenum.

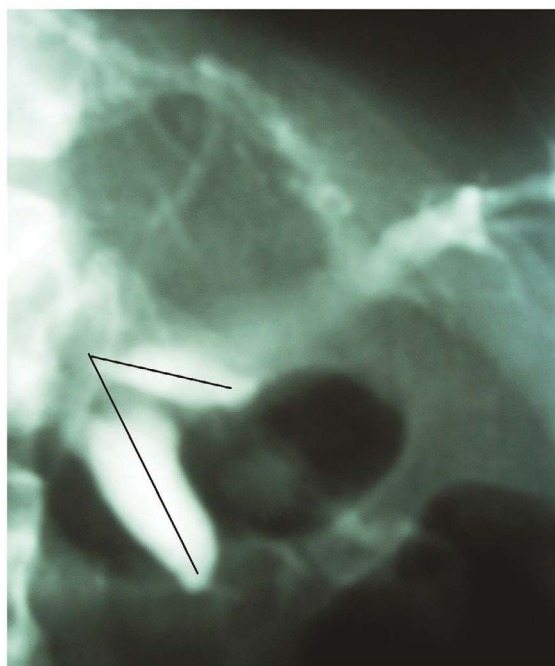


Figure 2: The image acquired immediately after the removal of the NBT revealed the presence of a pathological CBD angulation.

11 mm, with appropriate contrast in outflow to the duodenum (Figure 1). Given the patient's anamnesis, we decided to examine her biliary tree while she remained in a sitting position (the primary factor causing her symptoms). This examination was performed after introducing a nasobiliary tube (NBT); in particular, an X-ray image was obtained

after contrast injection into the CBD. The image acquired immediately after the removal of the NBT revealed the presence of a pathological CBD angulation, which explained the existing symptoms (Figure 2).

Discussion

Pathological CBD angulations may develop because of the coexistence of increased liver mobility and factors stabilizing the distal region of the CBD. In this situation, a change in body position (as described above) can cause varying degrees of CBD angulations. Predisposing factors for this phenomenon include postoperative adhesions after a prior cholecystectomy or choledochotomy; biliary T-tube drainage; intraoperative damage to the CBD (due to closure, ligation, and/or aggressive instrumental control); gastrointestinal, choledocho-intestinal or choledocho-choledochal (in cases involving liver transplantation) anastomosis; impact stones in the distal CBD; trauma; pancreatic tumors that displace the proximal CBD; pathological anatomical anomalies such as anomalous pancreaticobiliary duct union (APBDU); and congenital biliary stricture. Increased liver mobility has been observed in subjects with Marfan and Ehlers-Danlos syndromes [1,4-9]. In the present case, the most likely cause of severe CBD angulation was the development of adhesions after a previous cholecystectomy involving bile duct revision and T-tube biliary drainage.

Consequences

The existence of pathological CBD angulation may cause periodic mechanical jaundice and increase the risk of recurrent choledocholithiasis. These phenomena may begin to occur at 6 months after a successful biliary tree cleaning in approximately 5.2% (2.2%-12.5%) of patients [1,4]. Similar observations were noted by Keizman, who concluded that a CBD angle sharper than 145 degrees is an independent risk factor for recurrent stone formation after a successful endoscopic biliary tree cleaning [1,2]. Another risk factor is the fulfillment of Kim's criteria (a short distal arm and an angle sharper than 135 degrees). In patients with these risk factors, technical difficulties arise during the endoscopic removal of concretions due to an inability to appropriately place and adequately open a Dormia basket [1,10]. In the described patient, jaundice due to CBD angulation manifested only when the patient remained in a seated position for many hours while traveling by car. In contrast to the cases discussed above, we did not observe secondary choledocholithiasis; this lack of secondary choledocholithiasis can most likely be attributed to the infrequency with which the patient's symptoms appeared (one to two times per year).

Diagnosis

The most important diagnostic difficulty is that severe CBD angulation is temporary and dependent on body position. Most diagnostic tests (including ERCP performed under general anesthesia) evaluate CBD anatomy only in the supine position. Therefore, standard imaging studies only allow us to observe the indirect consequences of severe CBD angulation, such as recurrent choledocholithiasis without problems with CBD outflow after a successful biliary tree cleaning [9].

Although traditional ultrasound is a simple screening test, it exhibits particularly low sensitivity for early-stage occlusion and does not provide a three-dimensional image of the biliary tree [9]. Functional ultrasound after the ingestion of a fatty meal also has limited capacity with respect to diagnosing CBD angulation and can only provide indirect information regarding insufficient biliary outflow. Moreover, studies have indicated that this diagnostic tool performs well in cases involving

small bile ducts but fails in cases involving extended CBD and may produce false-negative results for up to 63.4% of patients in evaluations of biliary obstruction [11,12]. Nonetheless, the fact that these two types of ultrasound can be performed in different body positions creates certain opportunities for diagnosing pathological biliary angulation. Unfortunately, there is little information regarding this topic in the extant literature. In the present case, ultrasound examination revealed only a slight dilatation of the CBD without choledocholithiasis and was not helpful in establishing the patient's diagnosis.

Classic spiral CT cannot be used to visualize the biliary tract; computed tomography cholangiography (CTC), which has this capability, cannot be performed effectively if bilirubin levels are in excess of 2 mg/dl. In addition, the most important limitation restricting the detection of CBD angulation with this diagnostic tool is the fact that the examination must be performed in the supine position [13].

New equipment for magnetic resonance cholangiopancreatography (MRCP) can be used to provide a detailed, three-dimensional image of the biliary tree. However, this technique does not produce a dynamic picture and tends to overestimate lesion sizes [9,14]. In one study, MRCP was able to reveal all bile ducts in 55-82% of patients and detect narrowing in 76% of these patients; nonetheless, this technique cannot be utilized to assess the degree of obstruction. In addition, similarly to CT, MRCP is performed with the patient in the supine position, preventing the detection of severe CBD angulation in many cases [7,13,15-21].

Cholescintigraphy, similarly to CT and MRCP, is performed with the patient in the supine position; however, this technique also has minor significance as a diagnostic tool for assessing presenting pathology [18,22,23].

Due to its dynamic nature, ERCP can depict CBD angulation in certain patients [3]. In addition, if analgesedation is used to perform this procedure, the patient's position can be changed from prone to supine. This positional alteration increases the probability of detecting pathological CBD angulation. However, ERCP cannot visualize presenting pathology that appears only if the patient is in a vertical position (as in the described case). Therefore, we decided to utilize a different technique. After introducing an NBT, we shifted the patient to a sitting position; opacification of the biliary tree was then performed prior to obtaining a lateral X-ray. The picture acquired in this manner revealed an extremely sharp common bile duct angulation that explains the cause of the patient's symptoms. In the available literature, we have found no descriptions of the use of similar diagnostic techniques to diagnose CBD angulation.

Conclusion

In conclusion, it can be difficult to diagnose pathological CBD angulation because this problem is transient, occurring only when the body is in a specific position. Therefore, we draw attention to the possibility that this pathology may exist in patients for whom no source of symptoms can be identified by traditional ERCP examination. In addition, we observe that the use of NBT to opacify the bile ducts in a specific body position (based on a patient's medical anamnesis) can be a helpful tool for detecting CBD angulation.

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