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Paradoxical Gallbladder Distension Revealing an Aberrant Cystic Duct Insertion in Hilar Cholangiocarcinoma: A Case Report

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Abstract | **Case Report**

Anatomical variations of the cystic duct insertion are common and represent a major risk factor for complications in biliary surgery. We report the case of a 74-year-old woman admitted for an icteric cholestatic syndrome revealing a hilar cholangiocarcinoma, associated with paradoxical gallbladder distension. Imaging, particularly magnetic resonance cholangiopancreatography (MRCP), suggested an anatomical variation of the cystic duct that could explain this atypical presentation. A bilio-digestive bypass combined with cholecystectomy was performed. This case highlights the importance of preoperative recognition of cystic duct anatomical variations to ensure safe surgical management.

Keywords: Cystic duct; Anatomical variations; Cholestasis; cholangiocarcinoma; MRCP; Biliary surgery.

INTRODUCTION

The anatomy of the extrahepatic biliary tree is characterized by considerable variability, particularly regarding the cystic duct and its mode of insertion. Anatomical variations are frequent and may involve the course, level, and pattern of insertion of the cystic duct.

The advent of modern imaging techniques, especially magnetic resonance cholangiopancreatography (MRCP), has enabled better characterization of biliary anatomical variations and improved their preoperative identification. However, these anomalies may still be difficult to recognize, particularly in a tumoral context.

Through this case report, we illustrate a probable anatomical variation of the cystic duct insertion revealed by paradoxical gallbladder distension associated with a high biliary obstruction, and we discuss the diagnostic, therapeutic, and surgical implications of such variations.

PRESENTATION OF CASE

We report the case of a 74-year-old woman with no significant past medical history, admitted for a clinical cholestatic syndrome associated with generalized pruritus, evolving in an afebrile context with deterioration of the general condition.

On admission, clinical examination revealed a conscious patient who was hemodynamically and respiratory stable, presenting with diffuse cutaneomucosal jaundice and scratch lesions. Abdominal examination showed a soft, non-distended abdomen, without laparotomy scars, with a palpable gallbladder and no hepatomegaly, splenomegaly, or palpable abdominal mass.

Laboratory investigations demonstrated marked cholestasis with a total bilirubin level of 175.9 mg/L, including a conjugated bilirubin level of 97.7 mg/L, associated with elevated alkaline phosphatase (285 IU/L) and gamma-glutamyl transferase (229 IU/L), with moderate cytolysis (AST 94 IU/L, ALT 42 IU/L). Tumor markers were elevated, with carcinoembryonic antigen (CEA) at 11.7 ng/mL and CA 19-9 at 1786.3 IU/mL.

Abdominal ultrasound revealed marked dilatation of the intrahepatic bile ducts and the common bile duct with anechoic content, associated with a distended gallbladder containing a gallstone with posterior acoustic shadowing, as well as hilar hepatic lymphadenopathy, the largest measuring 10 mm in short-axis diameter.

Abdominopelvic computed tomography demonstrated a hydrocholecyst and dilatation of the intrahepatic bile ducts upstream of a tissue lesion at the biliary confluence, suggestive of cholangiocarcinoma.

This diagnosis was supported by MRCP, which showed an abrupt interruption of biliary flow at the level of the common bile duct confluence, with marked upstream dilatation of the intrahepatic bile ducts. The gallbladder was distended and lithiasic, while the cystic duct was poorly visualized, suggesting an anatomical variation that could explain gallbladder distension despite a high biliary obstruction, figure 1, figure 2.

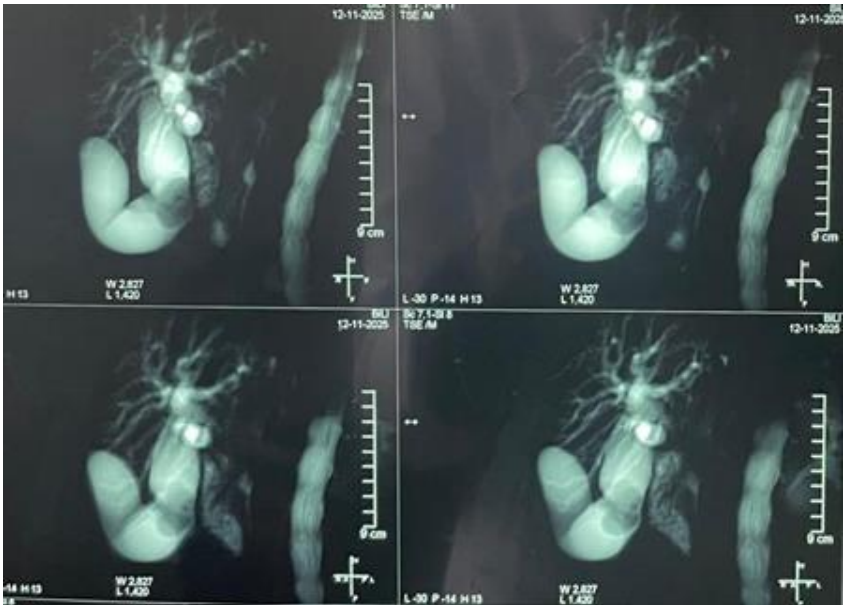

Figure 1: MRCP: abrupt interruption of biliary flow

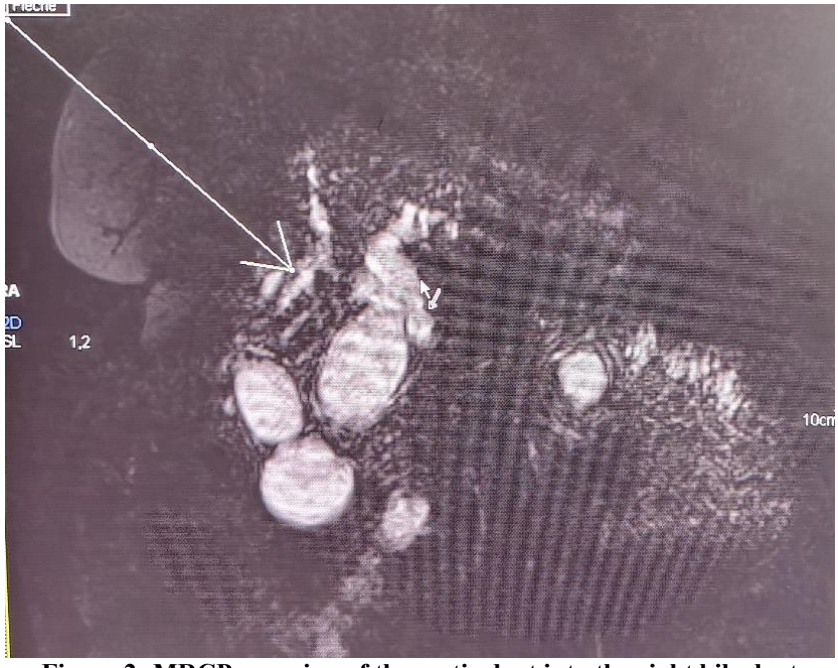

Figure 2: MRCP: opening of the cystic duct into the right bile duct

Thoracic CT scan revealed bilateral pulmonary nodules suspicious for secondary lesions. After multidisciplinary team discussion, surgical exploration was indicated, with the option of tumor resection or bilio-digestive bypass depending on intraoperative findings.

The abdominal cavity was approached through a right subcostal incision. Intraoperative exploration revealed no peritoneal effusion or hepatic or peritoneal nodules but showed an extensive tumor involving the primary biliary confluence, with posterior invasion of the portal vein trunk and a distended gallbladder wall harboring carcinomatosis nodules, and a cystic duct draining into the right hepatic duct.

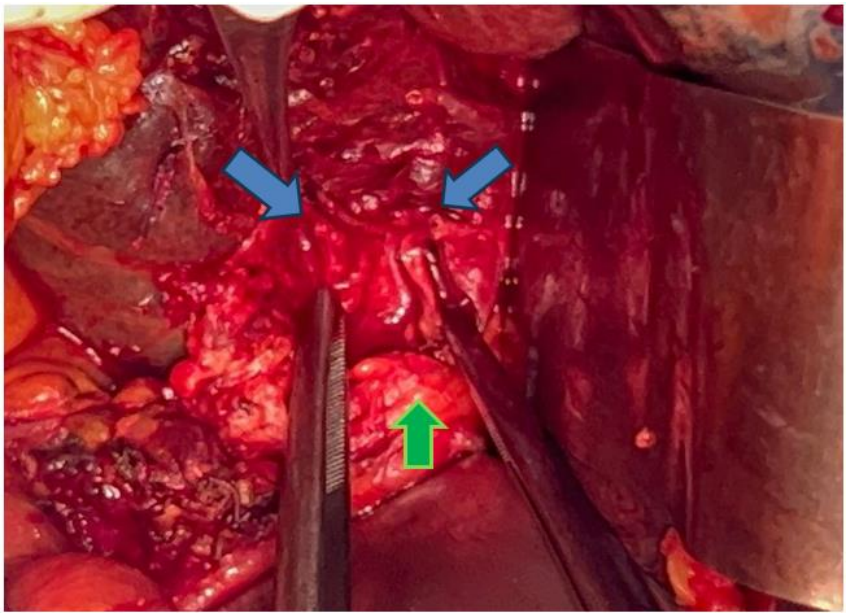

Figure 3: Intraoperative image showing the right and left hepatic ducts (blue arrow) after transection of the bile duct, and the tumor (green arrow)

The procedure consisted of an antegrade cholecystectomy with transection of the bile duct above the lesion, allowing the creation of a biliary neo-confluence, which was then anastomosed to a Roux-en-Y limb.

Opening of the bile duct allowed biopsy sampling, and frozen-section examination revealed a moderately differentiated invasive adenocarcinoma with perineural invasion pre- and retro-anastomotic drainage, completed by subhepatic and Douglas pouch drainage.

The postoperative course was marked, on postoperative day 3, by the development of a controlled biliary fistula, which was managed with antibiotic therapy consisting of ceftriaxone 2 g/day and metronidazole 500 mg, three time a day.

The outcome was favorable, with closure of the fistula after 18 days, after which the patient was discharged and referred to oncology for further management.

DISCUSSION

The anatomy of the cystic duct exhibits significant variability, which is recognized as a major determinant in the occurrence of complications in biliary surgery, particularly during laparoscopic cholecystectomy. In the so-called classical anatomical configuration, the cystic duct joins the lateral aspect of the common hepatic duct at the level of its middle third, contributing to the formation of the common bile duct. However, this standard arrangement is observed in only 40% to 80% of cases according to anatomical, radiological, and surgical series, highlighting the high prevalence of anatomical variations [1–3].

Advances in imaging, particularly magnetic resonance cholangiopancreatography (MRCP), have enabled detailed characterization of the various modes of cystic duct insertion. Several studies have shown that the classical lateral insertion accounts for approximately 50% to 77% of cases, while the remaining patients present atypical insertions, including medial, low, high, or posterior insertions [2,4,5]. A recent study demonstrated that nearly half of patients exhibited at least one anatomical variation of the cystic duct or the main bile duct, confirming that these anomalies are far from exceptional [6].

The main variants described in the literature include medial insertion of the cystic duct into the common bile duct, low insertion sometimes close to the ampulla of Vater, anterior or posterior spiral courses around the common bile duct, as well as high or aberrant insertions into the right hepatic duct or accessory sectoral ducts [1,4,5]. Certain rare variants, such as direct insertion of the cystic duct into the right hepatic duct or extremely low insertion, have been reported in less than 1% of cases, but are associated with a particularly high risk of iatrogenic bile duct injury when not identified [7,8].

From an embryological standpoint, these variations result from disturbances in the migration and differentiation of the hepatic and cystic buds during fetal development. Early separation may lead to a long cystic duct with an atypical course, whereas delayed separation may explain a short, absent, or abnormally inserted cystic duct [1].

Preoperative recognition of cystic duct anatomical variations is a key element of surgical planning. MRCP is currently considered the reference

non-invasive imaging modality for evaluation of the biliary tree, allowing precise visualization of intra- and extrahepatic bile ducts without the risks associated with invasive catheterization [4,6]. Several studies have demonstrated that MRCP enables identification of high-risk anatomical configurations, such as posterior or low insertions, and allows appropriate adaptation of the surgical strategy [4,9].

In a study assessing the impact of preoperative MRCP, anatomical variations were identified in approximately 26% of patients, with no major bile duct injuries reported when these variants were recognized prior to surgery [9]. These findings suggest that preoperative identification of anatomical anomalies may reduce the risk of bile duct injury, particularly in anatomically high-risk situations. Nevertheless, MRCP is not routinely indicated for all patients, but is strongly recommended in cases of suspected biliary anomalies or complex clinical settings.

The clinical consequences of cystic duct anatomical variations are substantial. Numerous studies have shown that intraoperative bile duct injuries most commonly occur in the setting of atypical anatomy, secondary to misidentification of structures within the hepatocystic (Calot's) triangle [10]. Confusion between the cystic duct and the common hepatic duct or an aberrant bile duct may result in inadvertent transection or ligation, leading to severe bile duct injuries requiring complex reparative procedures [10,11].

Associated complications include bile leaks, late biliary strictures, surgical reinterventions, prolonged hospital stay, and a significant increase in morbidity and mortality [10,11]. A recent observational study demonstrated that patients with cystic duct anatomical variations had a significantly higher incidence of intra- and postoperative complications compared with those presenting standard biliary anatomy [12].

In this context, the use of complementary intraoperative tools, such as intraoperative cholangiography or indocyanine green fluorescence imaging, is recommended when biliary anatomy is uncertain or atypical [11]. Thus, thorough knowledge of cystic duct anatomical variations, combined with adequate preoperative recognition and careful dissection respecting the principles of the "Critical View of Safety," remains essential to prevent complications in biliary surgery.

Conclusion

Anatomical variations of cystic duct insertion are common and represent a major challenge in daily surgical practice. Failure to recognize these variants exposes patients to an increased risk of iatrogenic bile duct injury, whereas their identification allows improved interpretation of atypical clinical presentations of biliary obstruction.

The reported case emphasizes the importance of considering a cystic duct anatomical anomaly when discordance exists between the level of biliary obstruction and gallbladder distension. MRCP plays a central role in the preoperative identification of these variants and in tailoring the surgical strategy.

Comprehensive knowledge of biliary anatomy, combined with meticulous imaging analysis and adherence to safe dissection principles, remains essential to optimize patient management and prevent complications in biliary surgery.

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Consent

As per international standard or university standard, patient(s) written consent has been collected and preserved by the author(s).

Ethical Approval

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

Conflicts Interests: Authors have declared that no competing interests exist.

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