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Variations of Biliary Tract and its Clinical Presentation

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Abstract

It is utmost important for the surgeons to have a comprehensive knowledge of the normal anatomy of the biliary system and its variations due to extraordinary frequency with which they perform in this anatomic site.

We report series of rare cases of congenital anomalies of biliary system diagnosed perioperatively. The first patient; a 32 year old female presented with features of obstructive jaundice and was found to have agenesis of the gallbladder at operation. This is a rare congenital malformation often associated with other anomalies. Agenesis of the gallbladder is difficult to diagnose clinically and discovery is often incidental during laparatomy for cholecystectomy. The second patient, a 67-year-old female, was electively admitted for laparoscopic cholecystectomy. Liver function tests showed a slightly deranged picture, CRP was elevated and the tumour marker, CA19.9 was raised CT abdomen and pelvis demonstrated acute- on- chronic cholecystitis? Gall bladder neoplasm. Initial laparoscopic cholecystectomy was converted to open laparatomy due to difficult anatomy and this revealed anomalous insertion of the cystic duct into right hepatic duct. Third patient 57 years female admitted with obstructive jaundice due to CBD stone ERCP failed to dislodge stone on cholecystectomy no cystic duct was found. Fourth patient 28 year old female admitted electively for laparoscopic cholecystectomy on operation found to have cystic artery arises from right hepatic crosses in front of common hepatic duct. These anomalies are important to recognize because great care must be taken to avoid inappropriate division of ducts and arteries in the course of cholecystectomy. Fifth patient 43-years old lady admitted through A/E with biliary colic slightly deranged LFTS with alkaline phosphatese 211 and GGT 319 U/S abdomen multiple gallstone CBD not dilated, liver normal at laparoscopic cholecystectomy accessory cystic duct (duct of luschika).

Conclusion: Most bile duct variations are asymptomatic, thus they may not be suspected pre-operatively without extensive pre-operative imaging. Thus, a difficult laparoscopic cholecystectomy requires prompt conversion to open laparotomy and/or intra-operative cholangiography to delineate the biliary tree and hence avoid injury.

Keywords: Cholecystectomy; Laparotomy, Gall bladder, Cholecystitis and cystic duct

Background and Objectives

Surgery on the biliary tract has posed a challenge for the surgeon. The anatomic anomalies in this area demand careful identification of the relevant structures. Apart from anatomic variations, there are congenital anomalies of biliary tree that arise from normal morphological development; variations in the division of the extra hepatic biliary ducts are very frequent and understandable by the sequence of embryological development of the biliary tree. Biliary system anomalies are common and are of variable clinical importance, an awareness of these variations in the biliary system is a potential factor in preventing ductal injury during surgery [1]. The goal of this study is to report these rare biliary variations detected during cholecystectomy, to underline the inadequacy of currently used diagnostic tests and the importance of arriving to a

correct preoperative diagnosis, in order to avoid needles surgical procedures and hence avoid potential complications.

Material and Methods

We report series of rare cases with congenital anomalies of the biliary system diagnosed intra-operatively.

First patient

A 32-year-old female patient was admitted via the accident and emergency department with a 4-day history of central abdominal pain, radiating to the back. The pain was worse at night, associated with anorexia, nausea, vomiting, diarrhoea and dark coloured urine. Clinically, the patient was jaundiced with an unremarkable abdominal examination. Biochemical analysis revealed deranged liver function tests (total bilirubin 55 iu/l, alanine transferase 204 iu/l, AST 98 iu/l, alkaline phosphatase 557 iu/l, albumin 45 g/l, amylase 13 iu/l), and in addition there was a moderate neutrophilia. Ultrasound of abdomen demonstrated grossly dilated intrahepatic ducts and common bile duct. The gallbladder was not visualised, but an acoustic shadow was visible in the gallbladder area, suspicious for a gallstone (**Figure 1**). Subsequent magnetic resonance cholangiopancreatogram (MRCP) similarly revealed marked dilatation of the intrahepatic biliary tree and significant dilatation of the upper common bile duct. Two filling defects were identified in the common bile duct, consistent with calculi, with a further four small calculi identified just above the ampulla. The gall bladder was not demonstrated and was presumed to be contracted secondary to disease process (**Figure 2**). The patient proceeded to endoscopic retrograde cholangiopancreatogram (ERCP), however this failed. This was repeated a few weeks later to completely clear the common bile duct at the first attempt (**Figure 3**).

The patient underwent an elective laparoscopic cholecystectomy10 weeks after initial presentation. The gallbladder was not identified during laparoscopy; therefore the procedure was converted to an open exploration of the biliary tract. Again, the gall bladder was not visible, leading to thorough exploration of all possible ectopic sites. The patient's postoperative recovery was uneventful and the patient was asymptomatic at follow up.

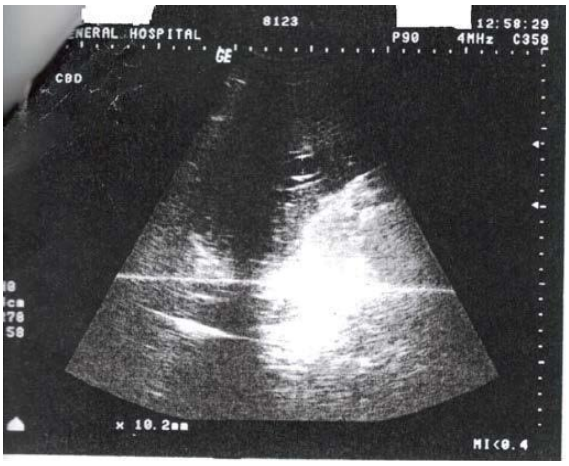

Figure 1 Acoustic shadow in the gall bladder area.

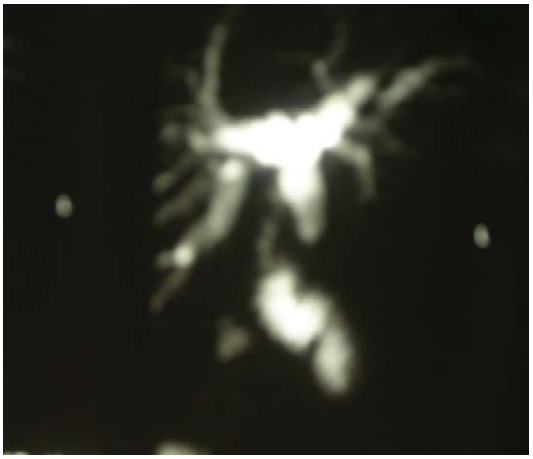

Figure 2 The gall bladder was not demonstrated and was presumed to be contracted secondary to disease process.

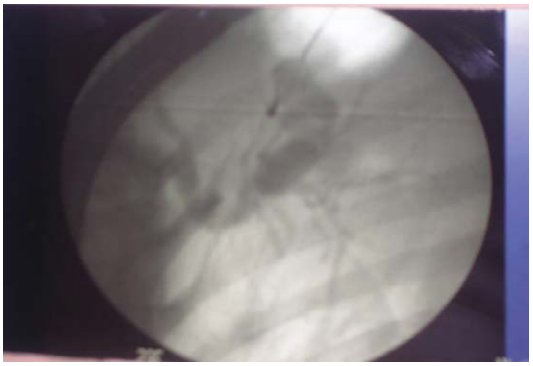

Figure 3 Few weeks later to completely clear the common bile duct at the first attempt.

Second patient

A 67-year old lady was admitted electively for laparoscopic cholecystectomy. She was followed in surgical OPD with diarrhoea and vomiting. There was incidental finding of hepatomegaly. Her full blood count was normal, LFT's showed a slightly deranged picture with normal bilirubin with an elevated GGT, ALP, and ALT. CRP was elevated at 33, tumour markers were also raised (CA 19.9). Abdominal ultrasound showed multiple gall stones with a normal gallbladder wall thickness (**Figure 4**). An abdomino-pelvic CT scan showed thick gallbladder wall, with pericholecystic fluid and fat stranding. A further CT was advised in six weeks to rule out a gallbladder malignancy (**Figure 5**). A Colonoscopy was normal. The patient underwent an elective laparoscopic cholecystectomy. The gall bladder was grossly distended with multiple dense adhesions. The anatomy of the Calot's triangle was grossly distorted. The laparoscopic procedure was abandoned and converted to an open procedure. On exploration, gall bladder wall and cystic duct were found to be thickened with multiple stones in the gallbladder. An intraoperative cholangiogram was performed. It demonstrated, the cystic duct emptying into the right hepatic duct rather than bile duct (**Figure 6**). The patient's post-operative recovery was uneventful.

Third patient

A 57 year old female was admitted through accident and

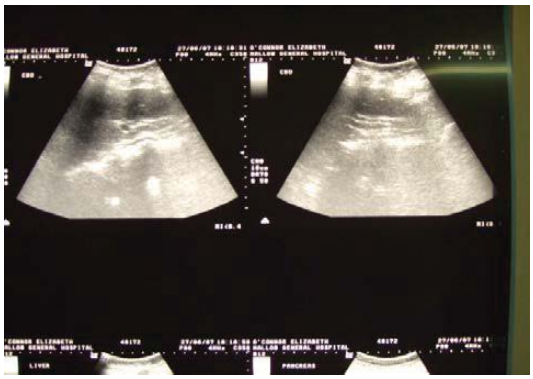

Figure 4 Abdominal ultrasound.

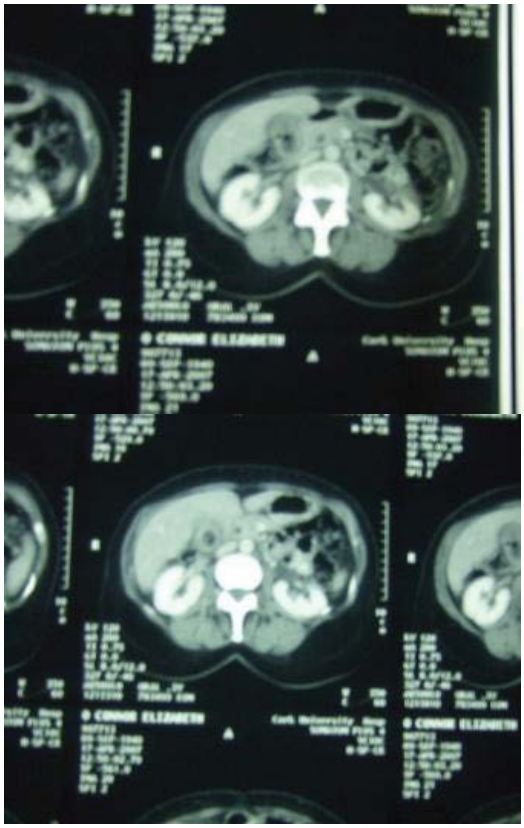

Figure 5 CT-Scan after six weeks..

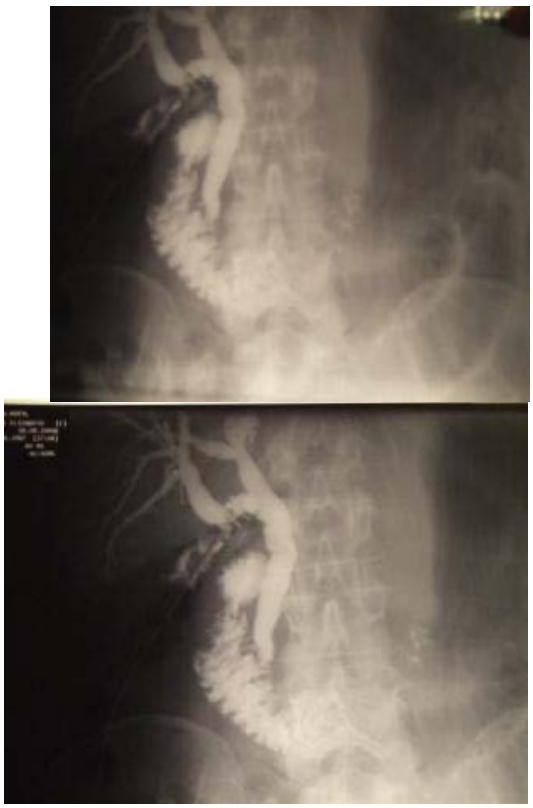

Figure 6 The cystic duct emptying [1].

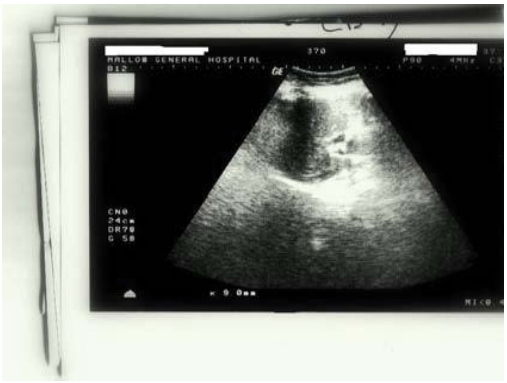

Figure 7 A further ERCP was performed several weeks later, but the stone.

emergency department with obstructive jaundice. She was clinically jaundiced with grossly deranged LFT's. An ERCP was performed, that demonstrated a stone impacted in common hepatic duct with copious amounts of pus in the common hepatic duct. A sphincterotomy was performed, but it failed to dislodge the stone and a stent was inserted. A further ERCP was performed several weeks later, but the stone could not be dislodged despite multiple attempts with mechanical lithotripsy and the stent was reinserted (**Figure 7**). An open cholecystectomy was performed. Operative cholangiogram revealed an absent cystic duct (**Figure 8**). Histology of the gallbladder showed an acute on chronic and Xanthogranulomatous cholecystitis.

Fourth patient

A 28 year old female was admitted for elective laparoscopic cholecystectomy. Routine blood investigations were all normal. U/S Abdomen showed a solitary gall stone with a normal CBD at laparoscopic cholecystectomy, it was noted that the cystic artery was arising from right hepatic artery and crossing in front of common hepatic duct. Histology of the gallbladder showed chronic cholecystitis.

Fifth patient

A 43 year old lady admitted through Accident and Emergency with biliary colic with slightly deranged LFTs with ALP of 211 and GGT of 319. Ultrasound of the abdomen showed multiple gallstones with CBD of normal diameter. At laparoscopic cholecystectomy accessory cystic duct (duct of Luschka) was

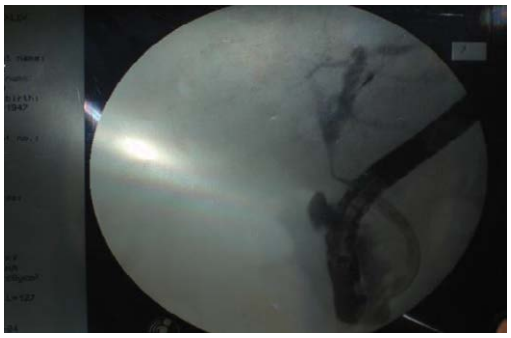

Figure 8 Operative cholangiogram revealed an absent cystic duct.

noted and clipped (**Figure 9**). Histology of the gallbladder shows chronic cholecystitis.

Discussion

A thorough knowledge of the biliary system and its potential variations is critical in preventing possible complications during laparoscopic cholecystectomy [2,3]. Anomalies of biliary duct may be problematic during surgical interventions, apart from iatrogenic injury to bile ducts during cholecystectomy, formation of bile duct calculi, pancreatitis, and cholangitis are other possible complications [4]. Failure of the development of cystic bud from the hepatic diverticulum results in agenesis of gallbladder and cystic duct. It occurs either as developmental phenomena or as an associated anomaly. Bergman in 1702 reported the concept of agenesis of gallbladder in humans [5]. So far only 413 cases have been reported in literature till July 2010 [6] (Figure 10). The incidence of gall bladder agenesis and cystic duct is extremely rare, ranging from 0.01% to 0.015%. An association of this entity with chromosomal abnormalities has been hypothesised. Bennion et al. formed a 3-group classification system for gall bladder agenesis based on a review of worldwide cases [7];

1. Gall bladder agenesis occurring in the setting of multiple foetal anomalies (15-16%) including cardiovascular, gastrointestinal and genitourinary.

2. Asymptomatic cases (35%) in which agenesis of gallbladder were found incidentally at autopsy or laparotomy. In some of the cases there was a familial association.

3. Symptomatic cases (50%) most commonly presenting with biliary colic (54%), dyspepsia (34%) and jaundice (27%). Agenesis of gallbladder poses a challenge to the surgeon during laparoscopic surgery whereby meticulous and thorough dissection of the entire hepatic biliary tree is essential in order to avoid inadvertent injury or excision of an anomalous cystic duct [8,9]. The diagnosis of gallbladder agenesis must be established intraoperatively as happened in our case [10]. The formal diagnosis of agenesis of the gallbladder requires a thorough examination of the entire abdominal cavity (retrohepatic, the left side of the abdomen, the lesser omentum, the falciform ligament, and the retro-peritoneum (retro-duodenal, retro-pancreatic) [11]. Anatomic variations of the cystic duct are usually of no clinical significance, occurring in 18-23% of cases, however unrecognized anatomic variations can be a source of confusion on imaging studies [12]. Huang et al.

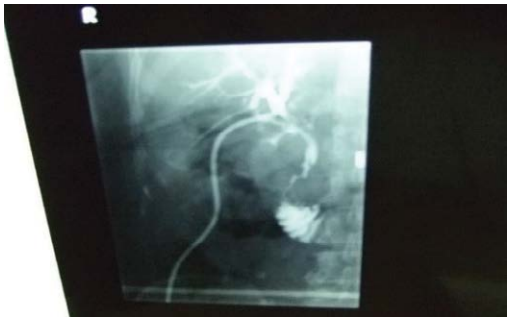

Figure 9 At laparoscopic cholecystectomy accessory cystic duct (duct of Luschka) was noted and clipped.

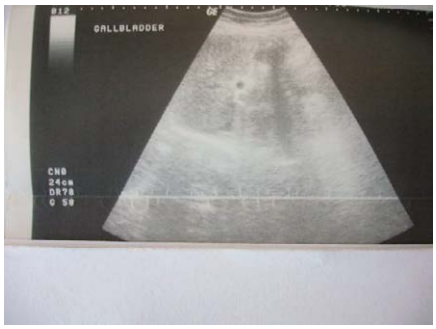

Figure 10 Ultrasound report-gall bladder.

reported anatomic variation of the confluence patterns of biliary tree using ERCP [13,14]. Anatomic variations in the cystic duct and hepatic duct are common, short or aberrant cystic duct entry or union with common hepatic duct is encountered routinely especially in the course of severe inflammation. However a congenital absence of the cystic duct is rare with severe surgical consequences. Anomalous insertion of the right hepatic duct into cystic duct is a rare anatomic variation [15]. Variations in the biliary ducts include accessory ducts, aberrant ducts which join right hepatic lobe directly with gallbladder (Cavalcanti et al. , Lamah et al., 2001, Foster and Wayson, 1962) The cystic duct has three common variations:

i) Course parallel to the common hepatic duct extending for 2 cm or more (1.5-25%) Turner and Fulcher (2001)

ii) Medial insertion through the left side of the common hepatic duct (10-17%);

iii) Insertion into the distal third of the common hepatic duct (9%);

On the other hand, insertion of the cystic duct into the right or left hepatic ducts, or influence of the left posterior hepatic duct directly into the cystic duct are considered rare (Turner and Fulcher; Yamakawa et al., 2007). A detailed anatomical knowledge of the possible variations in the anatomy of the cystic artery and its branches is critically important to the surgeon [16]. The cystic artery arising outside the hepatobiliary triangle usually passes ventral to the common hepatic duct and in some cases it may even be inferior to the cystic duct, thus becoming first structure encountered in dissection of the inferior border of

the hepatobiliary triangle and has a chance of accidental injury [17-19]. Anatomic variations of the cystic artery are commonly encountered, the course can also follow diverse paths, often in close proximity to the CBD (Price and Holden) [20-22], uncontrolled bleeding from the cystic artery and its branches pose a serious problem that may increase risk of damage to vital biliary and vascular structures [20,21]. Ducts of Luschka have gained increased clinical recognition in the era of laparoscopic cholecystectomy especially with advancement in technology and clear visualisation of the entire extra hepatic biliary system. The variability in their anatomic location puts them at risk during hepato biliary surgery [23]. They may not drain any liver parenchyma; but they can be a source of headache for a surgeon due to a persistent bile leak or rarely biliary peritonitis following hepatobiliary operations. If an accessory bile duct goes unrecognized at the time of the surgery, 5-7 days post operatively the patient will develop bile peritonitis, a complication that can be easily treated. However, if left untreated, morbidity as high as 44% has been reported. Recognition of biliary system anomalies are important, as great care must be taken to avoid inappropriate injury to the extra hepatic biliary ducts and arteries in the course of cholecystectomy. Use of imaging modalities e.g. intraoperative cholangiography may lead to an avoidance of iatrogenic bile duct injury [24]. Modern investigations (Ultrasonography, CT, ERCP, and NMR) have greatly improved the ability to image the biliary tract, however they still have limitations. The sensitivity of ultrasound though quite high ranges from 95-98% for diagnosis of cholelithiasis or common bile duct stones. Acoustic shadow misdiagnosed as stones can be due to artefacts or intestinal gas [7], subhepatic peritoneal folds [25,26], or periportal tissue [27]. Magnetic Resonance Cholangiogram is a non-invasive, and does not require contrast for visualisation of biliary tract. Preoperative MRCP should be considered mandatory in cases where ultrasonography has failed to visualise the gallbladder and a decision has been made for surgical intervention [28]. The use of sophisticated imaging modalities to identify biliary anomalies eg, intraoperative u/s, cholangioscintigraphy and selective arteriography may be useful but are only available in very specialised centres [29]. Despite excellent laparoscopic visualization anomalies of vascular structures, extra hepatic bile ducts are a common cause of intra and postoperative complications. Besides technical skills, experience and thorough knowledge of the anatomy of bile duct is required to decrease the incidence of post-operative biliary injuries. Cholangiography is mandatory whenever biliary anomalies are suspected during cholecystectomy [15]. Thus to summarise, this study highlights the fact that awareness of the variations in biliary tract and sound knowledge, clear dissection of all anatomic structures is vital before ligation, clipping and division in order to prevent duct injury, vascular injury, it is equally important to understand congenital variation of biliary and vascular anatomy as well as their operative implications [30].

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