

Residual Intrapancreatic Choledochal Cyst Following Incomplete Excision: A Case Report

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ABSTRACT

Choledochal Cysts (CCs) are rare, congenital anomalies characterised by dilatation of the biliary tract that predominantly affect young individuals. Surgical excision is the cornerstone of treatment; however, incomplete resection may predispose patients to serious complications, including recurrent cholangitis, pancreatitis, hepaticojejunostomy stricture, intrahepatic stone formation, and even malignant transformation. We report a 48-year-old woman who presented with a two-year history of recurrent upper abdominal pain and vomiting, ten years after excision of a CC with Roux-en-Y hepaticojejunostomy and sixteen years after an open cholecystectomy. Contrast-Enhanced Computed Tomography (CECT) demonstrated residual intrapancreatic CC containing calculi. At reoperation, the main portal vein was tracked, and a residual cyst extending into the intrapancreatic region was identified and completely excised, preserving the previous Roux-en-Y hepaticojejunostomy. The cyst contained pus and stones, and histopathological examination showed mucosal inflammation with hyperplasia, consistent with CCs. The postoperative course was uneventful, and the patient was discharged on postoperative day 6. She remained asymptomatic at six months of follow-up. This case underscores that complete excision of the intrapancreatic component at the index operation is essential to prevent long-term morbidity.

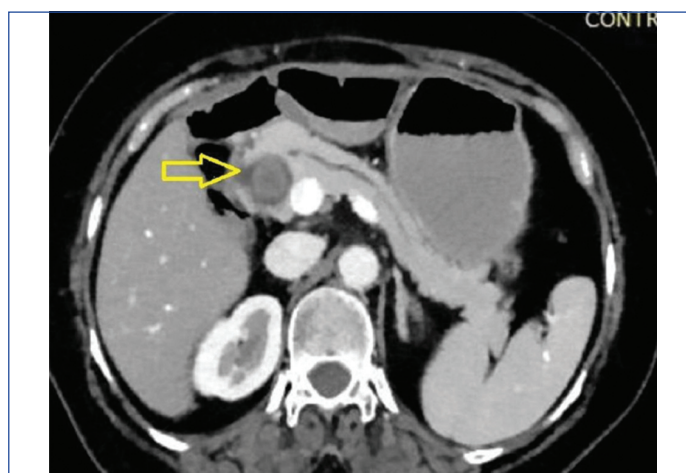
Keywords: Congenital biliary dilatation, Hepaticojejunostomy stricture, Intrahepatic stones, Recurrent cholangitis

CASE REPORT

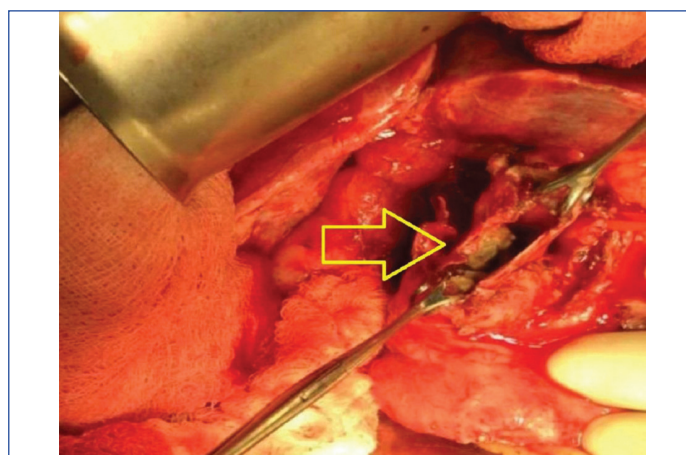
A 48-year-old female patient with no known comorbidities presented to the outpatient department with complaints of recurrent upper abdominal pain along with vomiting for the past two years. She had a past surgical history of an open cholecystectomy for symptomatic cholelithiasis at the age of 32 and excision of a CC with Roux-en-Y hepaticojejunostomy at the age of 38. On clinical examination, her vitals were stable and no icterus was noted. Per abdomen examination revealed epigastric tenderness with no other significant findings. Her preoperative investigations, including renal and liver function tests, were well within normal limits. CECT showed residual cystic dilatation of the terminal common bile duct within the pancreatic head, containing calculi. A diagnosis of residual CC was made [Table/Fig-1]. Endoscopic Retrograde Cholangiopancreatography (ERCP) was contemplated in view of the suspected intrapancreatic common bile duct debris, but was deliberately not undertaken: the surgically altered anatomy following the previous Roux-en-Y hepaticojejunostomy rendered endoscopic access to the papilla impractical, and the CECT findings were considered diagnostic and sufficient for operative planning.

The patient was taken up for open reoperation under general anaesthesia. A modified Makuuchi incision was made, and dense adhesions were noted. Adhesions were released, Kocherisation performed, and the residual cyst was identified. The pancreatic duct was in close relation and was carefully safeguarded. The cyst was excised, and the distal stump was transected and closed with 4-0 polydioxanone (PDS) suture. The previous hepaticojejunostomy was preserved. We placed a drain, and the operative time was about four hours with minimal blood loss.

Intraoperatively, we identified the main portal vein and found a residual intrapancreatic CC without disrupting the prior Roux-en-Y hepaticojejunostomy [Table/Fig-2]. The residual cyst, containing pus and stones, was successfully excised. The excised specimen measured 5×1.5 cm, with three small calculi (largest measuring 0.6×0.5 cm) and approximately 5 mL pus [Table/Fig-3]. Histopathological examination

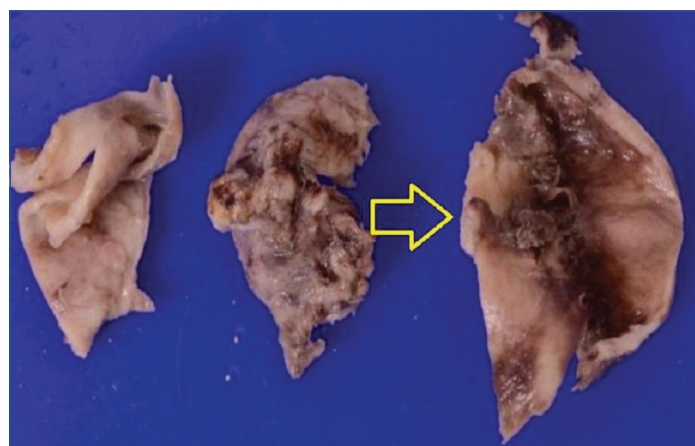


[Table/Fig-1]: Axial section, portal venous phase of preoperative Contrast-Enhanced Computed Tomography (CECT) image indicates the residual cystic dilatation of the terminal common bile duct within the pancreatic head. (yellow arrow).

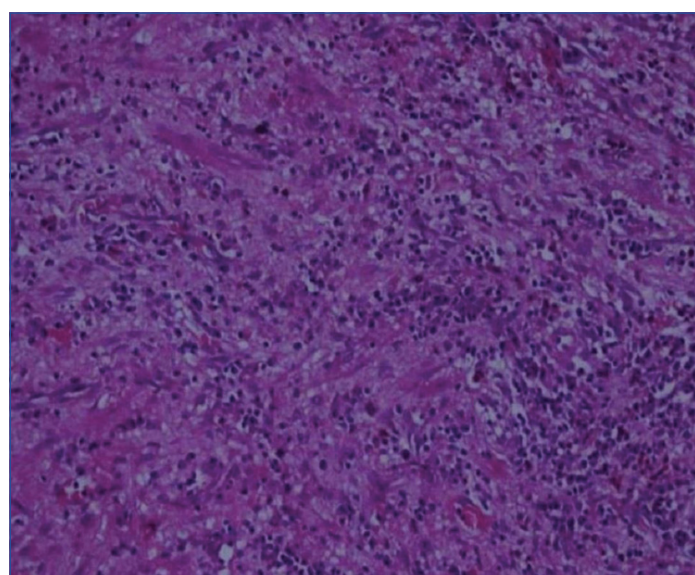


[Table/Fig-2]: Intraoperative imaging showing residual cyst loaded with stones. The yellow arrow indicates the opened residual cyst with calculi.

of the resected cyst wall revealed biliary tree epithelium with mucosal hyperplasia and an inflammatory infiltrate with a fibrocollagenous wall without any dysplasia [Table/Fig-4].



[Table/Fig-3]: Gross photograph of the excised residual Choledochal Cyst (CC) (5x1.5 cm). The yellow arrow indicates the laid-open cyst wall showing the inner mucosal surface.



[Table/Fig-4]: Haematoxylin and eosin stain, x100 magnification. The image shows dense chronic inflammatory infiltrates. There was no evidence of dysplasia or malignancy.

The patient's postoperative course was uneventful, with no complications. She was discharged on postoperative day six. At follow-up, she remained asymptomatic, with normal liver function tests and no radiological evidence of residual or recurrent disease at six months.

DISCUSSION

A CC is an aberrant dilatation of the biliary tract, most usually affecting the extrahepatic biliary tree. The distal extent of the dilatation may reach the intrapancreatic section of the bile duct [1]. CCs are rare medical conditions with an incidence in the Western population of 1 in 100 000-150 000 live births. The rate is remarkably higher in Asian populations, with a reported incidence of 1 in 1000, and about two-thirds of cases occur in Japan [1]. CC is most frequently observed in a female-to-male ratio of 4:1 [1].

When a CC extends deeply into the pancreas, complete excision is technically demanding and is often limited to avoid injuring the pancreatic duct - an injury that may precipitate recurrent pancreatitis [2]. The cyst wall left behind, however, is not inert: it retains its abnormal epithelial lining, remains exposed to refluxing pancreatic juice through the anomalous pancreaticobiliary junction, and drains poorly, so that chronic inflammation, bile stasis, and stone formation follow within the retained segment [3].

Although Roux-en-Y hepaticojejunostomy with complete excision of cyst is adequate for most type I, IV-B, and many IV-A CCs, anatomical variations may necessitate more complex and extensive surgeries [4]. Complete excision of the distal intrapancreatic component is essential and crucial; however, to avoid injury to the pancreatic duct and complications associated with it, the surgeon might leave a portion of the cyst wall at the distal end, which may lead to recurrence [5].

Our patient's course maps closely onto the largest reported series of remnant intrapancreatic cysts. Xia HT et al., reviewed 41 adults with remnant intrapancreatic CC after a primary excision; all experienced cholangitis, while cholangiolithiasis (66%), pancreatitis (51%) and vomiting (51%) were the next most frequent presentations, and the mean age at re-presentation was 49 years [5]. Xia HT et al., further reported an excellent or good outcome in 91% of patients after excision of the remnant cyst, which is consistent with the uneventful recovery and symptom-free six-month follow-up seen here. Their most sobering finding, however, was a 15% rate of malignant transformation, arising on average 140 months after the original incomplete excision [5]- an interval our patient had very nearly reached. Where anatomy or comorbidity precludes reoperation, Nguyen HV et al., have shown that combinations of minimally invasive endoscopic and percutaneous therapies can manage the complications of a retained cyst [6], but such measures address the sequelae rather than the retained epithelium itself. Therefore, complete surgical re-excision remains the definitive treatment.

Even after complete excision, late complications have been reported. A ten-year follow-up study documented biochemical liver dysfunction (10.71%), persistent intrahepatic bile duct dilatation (10.7%), recurrent abdominal pain (5.35%), and recurrence of common bile duct adenocarcinoma (1.78%) [7]. However, these rates were significantly lower than those observed following incomplete resection.

Various surgical approaches, such as the posterior pancreatic approach, the epicholedochal plexus approach, identifying the main portal vein, preoperative stenting, and the use of intraoperative cholangiography or ultrasonography, can aid in accomplishing a complete resection. Although ERCP is the gold standard for defining biliary anatomy, the non invasive Magnetic Resonance Cholangiopancreatography (MRCP) offers high-resolution imaging without the associated risks of instrumentation or radiation exposure [8].

CONCLUSION(S)

Leftover or residual CCs cannot only cause recurrence of symptoms, but also carry the risk of cholangiocarcinoma. Complete excision should be pursued whenever technically feasible, while avoiding injury to the pancreatic duct and adjacent structures.

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