



Complications in pediatric hepatobiliary surgery

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ABSTRACT

This review highlights the complications and their risk factors encountered in pediatric hepatobiliary surgery, specifically in the context of pediatric hepatic resection, excision of choledochal cyst, and the Kasai hepatoportoenterostomy procedure for biliary atresia as well as other procedures potentially affecting the biliary tree. With the understanding that these are relatively rare procedures, case reports and small case series are included in addition to larger series when available. The review focuses on publications in English over the past 15 years. Complications included both surgery-specific pathology, such as biliary stricture after excision of choledochal cyst, and disease-specific entities, such as malnutrition in biliary atresia. This review may be useful when considering a particular procedure or in the discussion thereof with a patient and family. Additionally, it illuminates the need for additional work with larger patient databases to refine and expand our knowledge of these complications and precipitating risk factors.

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Surgery of the hepatobiliary system remains crucially relevant to the field of pediatric surgery. This review will focus on complications from pediatric hepatic resection, excision of choledochal cyst, and the Kasai procedure for biliary atresia and will also briefly comment on other procedures that may impact the biliary tree. These complications are further divided into operative complications as compared to disease-specific complications. This review owes a great debt to Dr. Frederick Karrer's chapter entitled "Complications of Hepatobiliary Surgery" in the 2008 volume "Complications in Pediatric Surgery"¹; we largely sought to update the conclusions of that chapter with more recent data.

Hepatobiliary diseases such as choledochal cyst and biliary atresia are relatively uncommon with an incidence that is only a fraction of what is seen in other developmental anomalies. For instance, rates of congenital diaphragmatic hernia, tracheoesophageal fistula/esophageal atresia, and gastroschisis have been observed to be 1 per 2000–3000 live births; choledochal cysts may be as rare as 1 in 100,000 live births in Western countries, yet in some genetic groups in Asia and the Pacific are seen much more frequently.² Thus, as with many other entities in pediatric surgery, there often are insufficient data to make conclusive statements. For instance, there are no Cochrane Reviews addressing complications from any of the procedures that will be discussed in this review. Fortunately, the advent of a nationalized database, such as the recent merger of the "Biliary Atresia Research Consortium"

(BARC) with the "Cholestatic Liver Disease Consortium" (CLiC) to form the "Childhood Liver Disease Research Network" (ChiLDReN) in the United States has produced several relevant multicenter studies as have other national databases across the world. In this review, we will discuss the important considerations for complications after hepatic resection, operative intervention for choledochal cyst and surgical treatment for biliary atresia (Figure) as well as a brief section for procedures for acquired biliary pathology.

Hepatic Resection

Pediatric liver resections may be the rarest of the procedures discussed in this review and thus there is a paucity of data and even appropriately sized instruments³ for the surgeon. The lack of data prompts many surgeons to rely on studies in liver transplantation.⁴ Nearly 150 new cases of primary pediatric hepatic malignancy are diagnosed annually in the U.S. with two-thirds of these being hepatoblastoma and the rest comprised of hepatocellular carcinoma, sarcoma, and germ cell tumors with a roughly equal number of cases of metastatic disease of the liver, most often neuroblastoma, Wilms' tumor, and lymphoma.^{4,5} As curative resection is only possible in approximately 10% of primary liver tumors and up to 25% of metastatic liver tumors, additional approaches to increase rates of resectability have been proposed. Extensive resection can leave an infant or child with insufficient remaining liver parenchyma leading to postoperative liver failure.

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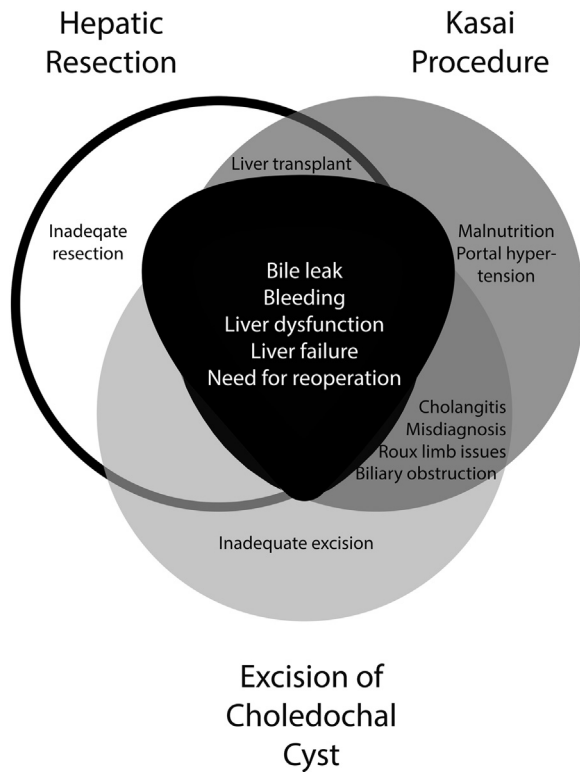


Fig. Venn diagram of complications seen in pediatric hepatobiliary surgery: hepatic resection, excision of choledochal cyst, and the Kasai procedure.

Wiederkehr et al⁶ recently described a technique for staged resection of marginally resectable liver tumors. Others have described “cherry-picking” lesions or performing nonanatomic resections⁷ to minimize loss of functional liver mass. In addition to hepatic insufficiency, other operation-specific complications of liver resection include biliary leak (potentially complicated by biliary cutaneous fistula⁸), intraoperative bleeding, cholangitis, and small bowel obstruction as compared to disease-specific complications like liver failure or necrosis with associated coagulopathy, recurrent disease, or eventual need for liver transplantation.

There is some information available on rates of complications and associated risk factors gleaned from small database studies. In 2009, a series of 126 patients undergoing liver resection for primary malignancy were identified from the Kids’ inpatient database (KID) and complications were assessed and compared based upon center volume. The average patient age was 5.8 years and the morbidity and mortality rates overall were 30.7% and 3.7%, respectively. A regression analysis revealed that surgery at a high volume center (≥ 5 resections per year) or need for blood transfusion increased the risk of postoperative complications. The former result is a surprising and counter-intuitive result but likely best explained by the authors’ inability to control for the extent of resection which was likely more significant at high volume centers. Indeed, the authors cautioned that high volume centers remain a valuable resource for patients with questionable resectability. This study, however, provides data and expectations regarding the overall outcomes and complications associated with pediatric liver resection (Table). Given the nature of this database study without the ability for follow-up, little complementary information is available to help understand the clinical impact of these patients’ complications.⁹

A prospective multinational study from Austria, Germany, and Switzerland evaluated the surgical data from 126 pediatric patients with hepatoblastoma; by nature of its design, it was able

Table

Complication, and Rates of Observation, After Liver Resection in a Series of 126 Patients.⁹

Complication	Rate Observed (%)
Digestive tract complication	7.4
Acute blood loss anemia	7.3
Respiratory complication	3.8
Urinary tract infection	3.5
Closure of wound dehiscence	1.3
Incisional hernia	1.3
Shock	1.3
Percutaneous drainage of abdominal collection	1.3
Other infection	1.3
Pneumonia	1.2
Central line infection	1.2
Re-exploration	1.1
Deep vein thrombosis	1.1

to provide additional information compared to the KID study about the impact of complications. The study found 15 patients (12%) with intraoperative complications and 26 (21%) with postoperative complications of whom 20 required reoperation. Intraoperative complications ranged from cardiac arrest due to air embolism to tumor rupture and severe hemorrhage (defined as blood loss greater than 50% of circulating volume). Postoperatively, they observed complications related to the recovery of liver synthetic function (liver failure, compromise in liver perfusion, and coagulopathy) and anatomic changes (bile leaks, cholestasis, adhesive small bowel obstruction, and eventration of the diaphragm). Complication rates were higher in patients with more advanced disease and extrahepatic tumor and often resulted in a delay in initiation of adjuvant chemotherapy, thus potentially contributing to the decrease in overall 5-year survival of these patients. Considering the severity and rates of complications, the authors advise evaluation for primary liver transplant in high risk cases.¹⁰

Even small studies though can help revise opinions about the feasibility of a surgical approach. Central hepatoblastoma presents a particularly difficult problem often only surgically treatable via a technically challenging central hepatectomy. This approach was initially thought to be associated with an increased risk of positive margins and with an increased risk of hepatic insufficiency compared to the option of an extended left or right hepatectomy but these concerns have not been supported by several recent case series. For instance, a French single center retrospective report of 9 patients, median age of 331 days, revealed the safety and efficacy of central hepatectomy.¹¹ A Spanish study of 5 patients, 2 with hepatoblastoma and 3 with other central liver tumors, had similar outcomes with adequate surgical margins and without postoperative liver failure.¹² However, extended left or right hepatic resection remains a viable surgical option: a report of 7 left extended hepatic resections, mean age of 3.1 years with a mean of 3.5 years of follow-up, revealed minimal surgical complications. The single patient with hepatocellular carcinoma died of recurrent disease 3.5 years after resection. There were no cases of biliary leak or stricture, no postoperative infections, no postoperative bleeding and all patients’ liver function normalized.¹³ There is not yet a direct comparison of extended hepatectomy and central hepatectomy for hepatoblastoma in the pediatric population but in the adult literature such a study showed that central hepatic resections were as safe, with perhaps less postoperative liver dysfunction, as extended hepatic resections.¹⁴

Pediatric hepatic resections may also be done for metastases. Addressing the risk/benefit ratio of hepatic metastectomy in the pediatric population, a single center series of 15 patients revealed no mortality and had 2 complications (bile leak and wound

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