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ORIGINAL ARTICLE

# Indications and outcomes of duodenum-preserving resection of the pancreatic head in children

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## Abstract

**Aim of study** Duodenum-preserving resection of the pancreatic head (DPRPH) with Roux-en-Y pancreatojejunostomy is a procedure used to remove focal pathological lesions of the pancreatic head. Although predominantly used in adult patients, it is both safe and effective in children. The aim of this study was to review our experience with this procedure, with focus on its indications, complications and long-term outcomes.

**Methods** A retrospective analysis of pediatric patients who underwent DPRPH between 1994 and 2015 was performed. Patient files were reviewed for demographic, diagnostic, operative and histological details, postoperative complications. Patients were contacted telephonically and sent questionnaires to determine long-term outcomes.

**Results** The study cohort consists of 21 patients, 14 girls and 7 boys, with an average age of 11.72 years (range 3 months to 18.6 years), who underwent DPRPH with end-to-end anastomosis of the jejunum to the pancreatic body (Roux-en-Y anastomosis). In four cases the head and also part of the body of the pancreas was resected. In the remaining 17 cases, only the head of the pancreas was resected. Indications for DPRPH were solid pseudopapillary tumor of the pancreas ($n=10$), trauma ($n=8$), pancreas divisum ($n=1$), focal congenital hyperinsulinism ($n=1$) and pancreatic cyst ($n=1$). The length of follow-up ranged from 1 to 22 years (average 9.66). One patient developed a biliary fistula, which closed spontaneously within 2 weeks after stent insertion. A recurrence of abdominal pain was reported in two patients, occurring at 7 months after the operation in one patient and at 1 year in the other. Pancreatic endocrine insufficiency did not occur in any of the 21 patients. Seven patients currently require a low fat diet, five of which need pancreatic enzyme supplementation. An additional two patients need enzyme supplementation without dietary restriction.

**Conclusion** DPRPH is a safe and effective procedure for the treatment of large focal pathological lesions of the pancreatic head in children. As a less invasive procedure than pancreatoduodenectomy, it is more appropriate for the developing child.

**Keywords** Pancreatic surgery · Duodenum-preserving resection of the pancreatic head · Children · Pancreatic insufficiency

## Introduction

Duodenum-preserving resection of the pancreatic head (DPRPH) has been used in adult surgery since 1972 [1]. The rationale to apply a duodenum-preserving resection in cases of benign inflammatory or cystic neoplastic lesions is to avoid resection of the gastric antrum and to ensure preservation of the duodenum and the extrahepatic biliary ducts [2]. The most common indication for DPRPH is chronic pancreatitis [3–6]. Less frequent indications include pancreas divisum with recurrent pancreatitis and abdominal pain [7, 8], solid pseudopapillary tumors of the pancreas (SPTP) [9–11] and trauma [12, 13]. DPRPH is a limited surgical procedure that offers the benefits of low early postoperative morbidity and conservation of metabolic functions in the long-term outcome of patients [14]. Quantitative comparison of DPRPH with pancreatoduodenectomy (PD) suggests that DPRPH, being a less invasive technique than PD, has benefits both in its short and long-term outcomes [15].

The aim of this study was to review our institutional experience with duodenum-preserving pancreatic head resection in children.

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## Patients and methods

A retrospective analysis was performed in children who underwent duodenum-preserving pancreatic head resection at our institution during a period of 22 years (January 1994–December 2015). We reviewed operative technique, histological diagnosis, complications and long-term results.

Duodenum-preserving resection of the pancreatic head is performed in two main steps. First, a total or subtotal resection of the head of the pancreas is performed while preserving the duodenum. In the second step, exocrine pancreatic secretory flow is restored by anastomosing the pancreatic duct with the first loop of jejunum.

After opening and exploring the abdominal cavity, the lesser sac is entered and the anterior aspect of the pancreatic head is fully exposed. The Kocher manoeuver is not necessary. After ligation and division of anterior superior and anterior inferior pancreaticoduodenal artery and vein (Fig. 1), resection of the head is performed with the use of an ultrasonic aspirator and dissector (in our case Cavitron ultrasonic surgical aspirator). The intrapancreatic portion of the distal common bile duct and origin of the posterior superior pancreaticoduodenal artery is identified and preserved (Fig. 2). The pancreatic duct is transected near the ampulla. Care has to be taken not to injure the bile duct. The body or tail of the pancreas is drained into the jejunum by means of a retrocolic single end-to-end, Roux-en-Y pancreaticojejunostomy.

Long-term outcome data were obtained by means of phone interview followed by a questionnaire sent in the mail. All contacted patients were asked to send stool samples for the assessment of human pancreatic elastase 1 concentration (detected by the sandwich ELISA method) and glycated hemoglobin levels (HbA1c, obtained by their general practitioners). Glycated hemoglobin levels were given in IFCC units and detected by immunoassay.

The questionnaire was sent to all 21 of our former patients. It consisted of nine items inquiring about their general health condition, any surgeries they subsequently underwent, hospital stays or out-patient visits due to related health problems (diarrhea, weight gain or loss, specific digestive problems), changes in bowel habits, problems with alimentation, allergies, medication, diabetes and pancreatitis. A total of 14 (67%) completed questionnaires were obtained.

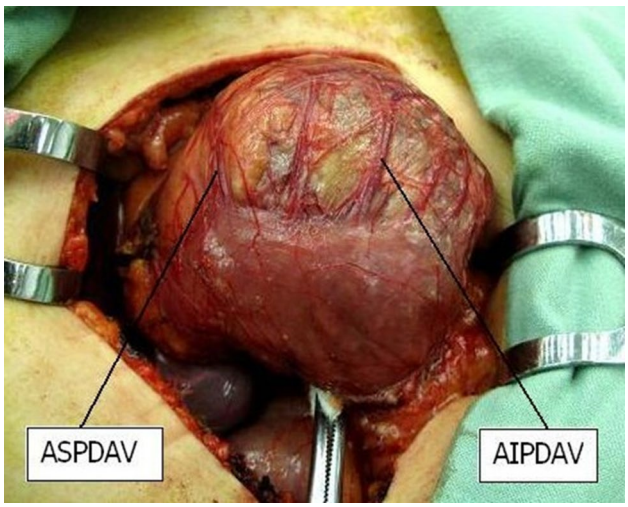


**Fig. 1** Tumor of the head of the pancreas with marked anterior superior and anterior inferior pancreaticoduodenal artery and vein (ASPDAV and AIPDAV)

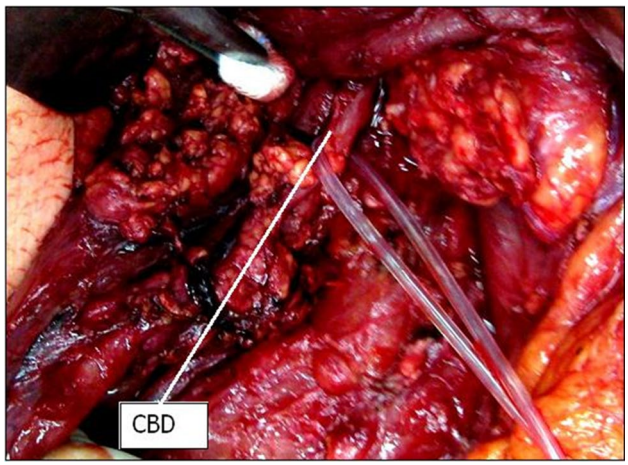


**Fig. 2** Identification of the distal common bile duct (CBD) during resection of the head of the pancreas

## Results

During the study period, DPRPH was performed in a total of 21 patients (14 girls and 7 boys). The average age at operation was 11.72 years (range 3 months–18.4 years). Table 1 shows the diagnosis, age at operation, sex and length of follow-up for each patient. The average duration of the procedure was 3.2 h (range 2.4–4.5 h). The average length of follow-up was 9.66 years (range 12 months–22 years).

One patient developed a biliary fistula, which closed after 2 weeks of stenting. In another patient, a serious hemorrhage from two gastric ulcers occurred 15 days after the operation. Attempt at endoscopic control of the hemorrhage failed and an urgent open operation was necessary to stop the bleeding. The patient was discharged 21 days after the second operation. Two patients complained of recurrent abdominal pain, occurring at 7 months postoperatively in one patient and at 1 year in the other. Over time, however, the pain subsided.

**Table 1** Overview of patients with performed duodenum-preserving resection of the pancreatic head—gender, age at the time of operation, diagnosis, location of the pathology and duration of follow-up

| No./sex | Age (years) | Diagnosis | Location | Follow-up |
|---|---|---|---|---|
| 1/f | 10.9 | SPTP | Head + body | 21 years AW |
| 2/f | 16.4 | SPTP | Head | 15.9 years AW |
| 3/m | 15 | SPTP | Head + body | 14.9 years AW |
| 4/f | 17.3 | SPTP | Head | 12.2 years AW |
| 5/f | 13.7 | SPTP | Head | 8 years AW |
| 6/f | 16.5 | SPTP | Head | 7.9 years AW |
| 7/f | 11.1 | SPTP | Head | 6.9 years AW |
| 8/f | 17.6 | SPTP | Head | 4.5 years AW |
| 9/f | 15.6 | SPTP | Head | 3.8 years AW |
| 10/f | 13 | SPTP | Head | 2.5 years AW |
| 11/m | 2.5 | Trauma | Head | 22 years AW |
| 12/f | 3 | Trauma | Head | 18.5 AW |
| 13/f | 12 | Trauma | Head | 16.2 years AW |
| 14/m | 11 | Trauma | Head | 15.4 years AW |
| 15/m | 17.5 | Trauma | Head | 7.1 AW |
| 16/f | 9.1 | Trauma | Head | 4.1 AW |
| 17/m | 11.6 | Trauma | Head | 4 years AWW |
| 18/m | 9.6 | Trauma | Head | 2.8 years AW |
| 19/f | 4 | PD | Head | 12.6 years AW |
| 20/m | 18.4 | Cyst | Head | 1.6 years AW |
| 21/m | 0.25 | FCH | Head | 1 year AW |

*f* female, *m* male, *SPTP* solid pseudopapillary tumor of the pancreas, *PD* pancreas divisum, *FCH* focal congenital hyperinsulinism, *AW* alive and well

A total of 67% (14 of total 21) of patients (or their mothers) responded to the questionnaires. Five patients from our questionnaire study group (36%) took medication regularly (pancreatic enzyme products, in two cases combined with omeprazole). Three patients underwent another surgery: ovarian cystectomy ($n=1$), appendectomy ($n=1$) and knee arthroscopy ($n=1$). None of these procedures were related to the primary diagnosis. Seven patients (50%) reported that they are on a diet (they avoided fatty and fried food) and seven patients (50%) eliminated specific irritant ingredients from their diet (garlic, onion, eggs, peppers, milk, carbonated drinks).

Five patients (36%) reported that they had been admitted to hospital for repeated attacks of pancreatitis, in most cases the problems receded after the first 2 years following the operation. None of the patients suffer from diabetes. Three declared some limitations in daily life (two patients cannot withstand strenuous work and one patient suffers from Crohn's disease and myasthenia gravis).

A total of 8 patients from the 14 that responded to the questionnaire sent stool samples and HbA1c results. None had elevated HbA1c levels and only two patients had low stool elastase levels, which prompted the commencement of pancreatic enzymes substitution therapy. However, they had not reported any problems with bowel habits (such as diarrhea, steatorrhea or atypical bowel habits).

We also obtained, with the patients' consent, medical information from local health care providers concerning the patients who had not answered the questionnaires. Only two of these patients have health complaints: one boy is treated for perianal complications of Crohn's disease and another patient is undergoing treatment for depression following a suicide attempt. According to the patient's psychiatrist, the cause is unrelated to the pancreatic operation.

At the time of completion of our study, seven patients require some level of pancreatic exocrine substitution therapy (33% of the total patient cohort).

## Discussion

### Solid pseudopapillary tumor of the pancreas

A solid pseudopapillary tumor of the pancreas (SPTP) is a rare pancreatic tumor that accounts for only 2–3% of all primary malignant pancreatic tumors [16]. It was first described by Frantz in 1959 [17] and was defined as a solid pseudopapillary tumor of the pancreas by the World Health Organization in 1996. It is of low malignant potential, and radical resection offers an excellent prognosis. However, local recurrence and distant metastasis after incomplete resection have been reported. Because of the rarity of SPTP in children, surgical intervention strategies are still controversial. The operative procedures range from tumor enucleation to distal pancreatectomy and pancreatico-duodenectomy [18].

Most pediatric surgeons prefer pancreatico-duodenectomy or modified a Whipple procedure for tumors located in the head of the pancreas [19–25]. In 2011 Zampieri et al. presented five female children with SPTP located in the head of the pancreas treated with duodenum-preserving pancreatic head resection [10]. Resende at al. presented a 16-year-old female patient with abdominal pain and dyspepsia. After a radiological diagnosis of SPTP was established the patient underwent DPRPH [26]. In our department, we have been conducting DPRPH in patients with SPTP since 1995 [27].

### Pancreatic trauma

Pancreatic trauma is rare in children, and management strategies are diverse and controversial. Grade I and II injuries constitute the majority of pancreatic trauma. Drainage with any required local debridement is most commonly performed. Grade III injuries generally require distal pancreatectomy and splenectomy with drainage. In children, an effort should be made to preserve the spleen if possible because of the potential for overwhelming post-splenectomy

infection. The management of grade IV and grade V injury is complex, requiring the input of experienced hepatopancreatobiliary surgeons. If the duodenum is not compromised and the ampulla is intact, the most straightforward option is washout and drainage, although pancreatic head resection may result in fewer subsequent operations and complications. In cases of massive pancreatic head disruption, pancreaticoduodenectomy may be required [28].

Pancreatic trauma may be managed non-operatively or may require early surgical intervention. Early operative intervention is associated with shorter lengths of hospitalization and decreased morbidity when compared with non-operative management. Furthermore, non-operative management of complete pancreatic transection has been associated with longer duration of total parenteral nutrition (TPN), recurrent bouts of pancreatitis, eventual surgical intervention, family inconvenience and stress, and increased overall hospital costs. Borkon [29] recommends early operative intervention for all complete pancreatic transections in children sustaining blunt abdominal trauma, with the specific procedure individualized. In pancreatic head trauma type IV he recommends Roux-en-Y distal pancreaticojejunostomy (RYPJ). Beres advocates early surgical intervention for patients with ductal transection if feasible. The primary advantages of surgery are likely restricted to those patients presenting acutely with injury amenable to resection [30]. Stringer concluded that children referred early with clearly defined grade III injuries probably benefit from an early spleen-sparing distal pancreatectomy. Those with grade IV injuries frequently require laparotomy when Roux-en-Y drainage of the fracture site is a useful technique [31]. In adult patients, DPRPH is extremely rarely performed [13, 32]. In our institute we have been conducting DPRPH since 1994 for cases of pancreatic trauma [12].

## Pancreas divisum

Pancreas divisum can cause recurrent pancreatitis and chronic abdominal pain. Patients with symptomatic PD are treated by ERCP papillotomy. If there is no improvement after ERCP or if ERCP is not technically feasible then surgical intervention is indicated. Surgery in pancreas divisum is very rarely performed and is only considered in patients with repeated pancreatitis, chronic abdominal pain and failure to thrive [33].

O'Rourke reported a case of an 8-year-old girl who presented with recurrent bouts of acute pancreatitis and multiple failed attempts at endoscopic retrograde cholangiopancreatography (ERCP) who indicated for surgical exploration. She underwent open sphincteroplasty of both major and minor papillae and remained symptom-free for a follow-up period of 22 months [34]. Neblet et al. identified pancreas divisum in 10 children (19.2% of children with relapsing or chronic pancreatitis). Seven underwent transduodenal sphincteroplasty of the accessory papilla and three patients underwent longitudinal pancreaticojejunostomy. Of the three patients who underwent pancreaticojejunostomy, symptoms remarkably improved in two [35].

Rabinovich et al. presented the largest series in the pediatric literature of pancreatic procedures performed in children. Over a 12 years period, they performed 72 pancreatic operations in 62 pediatric patients. Nine patients underwent operative treatment for pancreas divisum causing repeated bouts of pancreatitis. These patients were treated with the Frey and Puestow procedures. Long-term follow-up of these patients was not reported [36].

DPRPH is an alternative to other resections or drainage procedures after the failure of endoscopic treatment. The first report of pancreas divisum in adult patients treated by DPRPH was written by the Beger group in Ulm [37]. In their paper, the authors describe 36 patients with pancreatitis caused by PD who were treated by DPRPH. Following surgery, 50% of the patients were completely pain-free and 31% reported a significant reduction of pain.

We performed DPRPH in a child with pancreas divisum in 2014, which, to our knowledge, is the first published case [38].

## Focal congenital hypersplenism

Congenital hyperinsulinism of infancy (CHI) is characterized by excessive insulin secretion resulting in persistent hypoglycemia, which can lead to irreversible severe neurological damage in infants. Near-total pancreatectomy is the procedure of choice for diffuse CHI, whereas a localized resection is curative in focal CHI [39].

In 2012 Laje et al. published a report of 23 patients with focal CHI who underwent pancreatic head resection and Roux-en-Y pancreaticojejunostomy. Twenty-one patients underwent near-total pancreatic head resection and two patients underwent pylorus-preserving Whipple procedures. The authors concluded that pancreatic head resection with Roux-en-Y pancreaticojejunostomy is a safe and effective procedure for the treatment of CHI in patients with large focal lesions of the pancreatic head non-amenable to local resection alone [40]. Obatake et al. [41] presented a case of a 2-month-old boy, who presented with uncontrollable persistent hypoglycemia and was subsequently diagnosed with focal CHI of the pancreatic head and treated by pancreatic head resection preserving the main pancreatic duct.

## Congenital cysts

Congenital true pancreatic cysts, which are not the result of trauma, are rare and represent less than 1% of all pancreatic cysts in children [42]. The exact embryological origin of

congenital pancreatic cysts is unknown. They are believed to arise from a developmental anomaly of the pancreatic ductal system. The treatment of congenital pancreatic cyst is surgical. The type of surgical treatment depends on the location and size of the cyst. Total excision is the treatment of choice. This is usually feasible for cysts located in the body and tail, which may necessitate distal pancreatectomy. Cysts located in the head of the pancreas are difficult to excise totally and are better managed via internal drainage via either cystoduodenostomy or Roux-en-Y cystojejunostomy [43].

### Postoperative endocrine and exocrine pancreatic insufficiency

The most frequent long-term complications of pancreatic surgery in both children and adult patients are exocrine and endocrine dysfunction of the pancreas, gastrointestinal dysfunction (including delayed gastric emptying, abdominal pain and change in quality and frequency of stools), impaired quality of life, persisting impairment of nutritional status, fatigue, and local surgical-site related complications (including pancreatic fistula or bleeding) [44–52]. Routine monitoring of elastase levels, as well as fasting glucose or HbA1c levels in blood is not common during long-term follow-up, but may be of benefit for patients who have undergone pancreatic surgery.

## Conclusions

Duodenum-preserving resection of the pancreatic head with Roux-en-Y pancreaticojejunostomy is a safe and effective procedure for the treatment of large focal lesions of the pancreatic head in children. The authors consider DPHRP technique more appropriate for the developing organism of a child. They are convinced that the reported favorable short and long-term results support their view.

### Compliance with ethical standards

**Conflict of interest** All authors declare that they have no conflicts of interest.

**Ethical approval** All procedures performed were in accordance with the ethical standards of the institutions involved, in accordance with national ethical standards and with the 1964 Helsinki declaration.

**Informed consent** Informed consent was obtained from all individual participants included in the study.

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