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Laparoscopic resection of pancreatic neck lesion with Roux-en-Y pancreatico-jejunostomy

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A B S T R A C T

Background: Congenital hyperinsulinism is a rare disease and patients not responding to medical treatment need near-total or partial pancreatectomy, dependent on whether they have diffuse or focal hyperinsulinism, respectively. While laparoscopic technique for distal and for total pancreatectomy has been developed, minimally invasive resection of the pancreatic neck with pancreatico-jejunostomy has not been reported in children before.

Case summary: A 2-year old boy suffered from congenital hyperinsulinism, which was refractory to high-dose medical treatment. The nuclear-medicine scan revealed a focal lesion of the pancreatic neck, hence partial pancreatectomy was indicated. On laparoscopy, a slightly prominent tissue mass was apparent in the area of the pancreatic neck. We proceeded with laparoscopic mobilisation of the pancreas from the underlying splenic vessels and resected the pancreatic neck and adjacent parts of the body and the head. After macroscopic resection of the mass, the patient's intraoperative blood glucose levels increased to a point where insulin had to be substituted. To drain the pancreatic tail, we formed an end-to-side anastomosis in the proximal Jejunum and brought the open end to the pancreatic tail and performed a laparoscopic pancreatico-jejunostomy. The patient tolerated the procedure well and had no remaining signs or symptoms of hyperinsulinism.

Conclusion: This is the first report of a laparoscopic middle-segmental pancreatic resection with laparoscopic assisted Roux-en-Y pancreatico-jejunostomy in a child. For benign pancreatic lesions proximal to the body and tail, the described minimally invasive technique should be considered.

1. Introduction

Congenital hyperinsulinism (HI) occurs in 1–1.4 neonates of 50′000 live births [1–3]. However, it is more common up to 1 in 2500 in consanguineous populations. Patients who do not respond to medical treatment require partial or near-total pancreatectomy for focal or diffuse HI, respectively [4]. While open surgical technique is still the most commonly chosen approach for subtotal pancreatectomy in cases of diffuse HI, minimally invasive surgery (MIS) is an alternate approach for focal lesions, especially when they are located in the pancreatic tail [5]. Imaging studies combining data from computed tomography and nuclear medical scans can be particularly helpful in preoperative planning in focal disease [6,7]. Intraoperative frozen sections are indispensable for confirming the diagnosis and determining the extent of resection for both, diffuse and focal forms of HI. Herein, we present a case of focal congenital HI, with challenging intraoperative decision making and the first description of a paediatric minimally invasive resection of the pancreatic neck and body with laparoscopic pancreatico-jejunostomy.

2. Case report

A 2-year old boy with congenital HI with paternal *ABCC8* mutation had intermittent hypoglycaemic episodes despite being on intensive feeding regime (3-hourly bolus feeds and overnight continuous feeds via gastrostomy), high-dose treatment with Octreotide at 39 µg/kg/day and Sirolimus 0.8 mg twice daily (3.2 mg/m^2/day). The child had maximum fasting tolerance of 3 h. A focal lesion of the pancreatic neck was confirmed on a 18F-DOPA PET scan done 4 weeks prior to the planned operation (Fig. 1). A peripherally inserted central catheter (PICC) was placed 3 days preoperatively.

After induction of anaesthesia a single dose of Co-Amoxicillin was administered, and an epidural catheter was placed. We placed a 10 mm Hasson port supra umbilically and working ports in both flanks. After entering the lesser sac through the gastrocolic ligament, we placed a Nathanson liver retractor to elevate the stomach, which provided access to the pancreas. We identified a tissue mass in the neck area (Fig. 1). Freeing up the cranial border of the pancreas using hook cautery followed by separating the pancreas from the splenic vessels starting at the lower border of the pancreatic body allowed complete transection of the pancreatic body distal to the perceived nodular mass (Fig. 2). We

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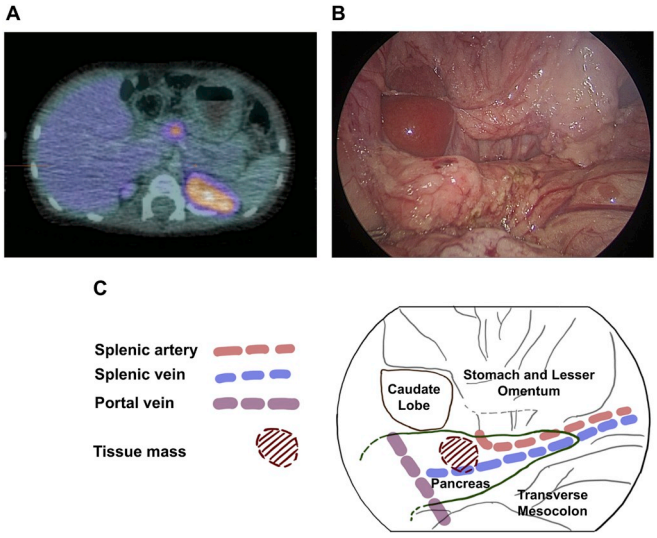

Fig. 1. A) Axial image of an ^{18}F-DOPA PET contrast enhanced CT scan at the level of the pancreas, showing a focal uptake in the area of the pancreatic neck. (Abbreviations: ^{18}F-DOPA = 6-[18F]-L-fluoro-L-3, 4-dihydroxyphenylalanine. PET = positron emission tomography).
B) Laparoscopic view after entering the lesser sac through the gastrocolic ligament and elevating the stomach using a Nathanson retractor. A slightly prominent tissue mass is visible at the pancreatic neck.
C) Schematic drawing of laparoscopic findings. The major vessels which the pancreas is overlying are shown.

then continued the separation of the pancreas from the splenic vessels towards the neck until the mass with the adjacent normal appearing pancreas could be removed via the umbilical port. The proximal transection of the pancreas was carried out using a laparoscopic Harmonic® (Ethicon®, Cincinnati, OH) scalpel to seal the parenchyma and the pancreatic duct (Fig. 3), while laparoscopic hook cautery was used for the previous distal transection [8].

Macroscopically, the transected mass had a fibrous appearance and measured 6 mm by 7 mm (Fig. 3). On Hematoxylin-Eosin (HE)-stained frozen sections, there were sporadic islet cell hypertrophy without the clear presence of enlarged islets which made it difficult to confirm the lesion was completely removed at frozen section; only the immunohistochemical stains done the following days showed the typical changes of a focal lesion responsible for HI (Fig. 3). Therefore, we carried out the dissection further first towards the pancreatic tail and then towards the head to remove two additional specimens (Fig. 4), which both showed normal macroscopic appearance as well as normal microscopic structure on HE-stained frozen sections. Following removal of the mass, the patient's blood glucose levels started to increase intraoperatively and reached a peak of 17.4 mmol/L on a glucose infusion rate of 5 mg/kg/min, requiring insulin infusion. Despite the lack of histologic confirmation of removal of the lesion, we considered it substantively resected based on the macroscopic appearance, the preliminary finding of the frozen sections, and on the hyperglycaemic response. As the preserved pancreatic tail was more than 5 cm long, we decided to preserve it and to assure drainage with a Roux-en-Y pancreatico-jejunostomy. To this end, we identified the proximal jejunum, brought it outside the abdomen through a short extension of the umbilical incision [9]. We subsequently formed a foot-point anastomosis, dividing the Jejunum 30 cm from the ligament of Treitz with subsequent end-to-side (antimesenteric) anastomosis using a 5-0 absorbable monofilament suture in a running fashion (Fig. 4). Intraabdominally again, we then brought the Roux jejunal limb through the transverse mesocolon to the pancreatic tail, which was thoroughly tucked into the open end of the Jejunum (Figs. 4 and 5); the pancreatico-jejunal anastomosis was carried out using eight interrupted stitches of 5-0 PDS® (Ethicon®, Cincinnati, OH; Fig. 5) and the hiatus created in the transverse mesocolon was closed around the jejunal Roux limb. An 8-French silicone rubber drain was placed in the lesser sac before closure of all port sites. Duration of the procedure was 210 min, which included awaiting the results of frozen sections three times.

Postoperatively, the insulin infusion could be weaned off and stopped after 6 h – he later reached a fasting tolerance of 12 h without developing hypoglycaemia and undetectable insulin with generation of fatty acids and ketones at the end of the fast suggestive of complete resolution of HI. Diagnostic imaging performed to investigate recurrent fever spikes and an elevated CRP (up to 165 μmol/L) revealed a multiloculated collection adjacent to the left lobe of the liver, which was successfully treated with a 3 weeks course of intravenous antibiotics. Besides the fever spikes, the patient recovered well from the procedure and full oral feeds were reached after 9 days. Drain output, which contained pancreatic secretions as confirmed by high amylase levels, subsided 10 days after the procedure. Total inpatient care after the operation was 4 weeks due to the course of intravenous antibiotic treatment, which was extended to 3 weeks in the context of previous long-standing immunosuppressive treatment. A recent ultrasound showed complete resolution of the collection and no pancreatic duct dilatations.

3. Discussion

Congenital HI is arguably the most common disease requiring pancreatic resection in infants and toddlers, other indications such as pancreatic neoplasms or chronic pancreatitis are exceedingly rare in this age group. The type of disease determines the extent of pancreatic resection; in focal disease, only as much tissue is resected as to safely remove the pathologic mass.

Open operation is still the most common approach for near-total pancreatectomy in cases of diffuse HI [10,11], although the technique of laparoscopic near-total pancreatectomy has been developed and described [5,12]. The open approach in general has several advantages, it does not only allow for palpation of the pancreas for masses or nodules, it also permits easier access for intraoperative ultrasound than laparoscopy. Intraoperative ultrasound is often essential to locate an insulinoma and to avoid injury to the pancreatic duct [13–16]. On the other hand, there are several reports of laparoscopic pancreatic resections, especially with focal lesions within the pancreatic tail [5,12,17,18]. Surgeons may choose a laparoscopic approach for its reduced morbidity and pain and avoidance of the undesirable cosmetic result of an upper transverse laparotomy; some also report much earlier oral feeding and shorter hospitalization after laparoscopic resection compared to open [18–20]. The use of intraoperative ultrasound is also recommended for laparoscopic resections [15,19]. However, laparoscopic ultrasound may not be available in a paediatric hospital. Furthermore, some reports raise the concern of an increased likelihood of incomplete resection and the subsequent need for re-operation after laparoscopic pancreatectomy [21]. Leakage of pancreatic fluid and fistula formation is an additional concern with minimally invasive pancreatectomy and pancreatico-jejunostomy [22]. In our case, output from the drain placed in the lesser sac decreased over time and subsided spontaneously on postoperative day 10. Overall, minimally invasive pancreatectomy has important limitations; yet, successful laparoscopic operations also offers distinct benefits to the patient.

In case of focal HI – such as in our patient – the mass can frequently be enucleated resulting in a less than 5% loss of pancreatic tissue [16]. However, some patients with head or neck lesions of the pancreas still require a proximal pancreatectomy and thus a pancreatico-jejunostomy; additionally, enucleation can lead to injury of the pancreatic duct [14,16]. In our case, at the point where we were confident that we had removed the lesion, we had three options. First, to remove the remaining tail, leaving less than 50% of pancreatic tissue having removed already some of the head of the pancreas with the long-term risk

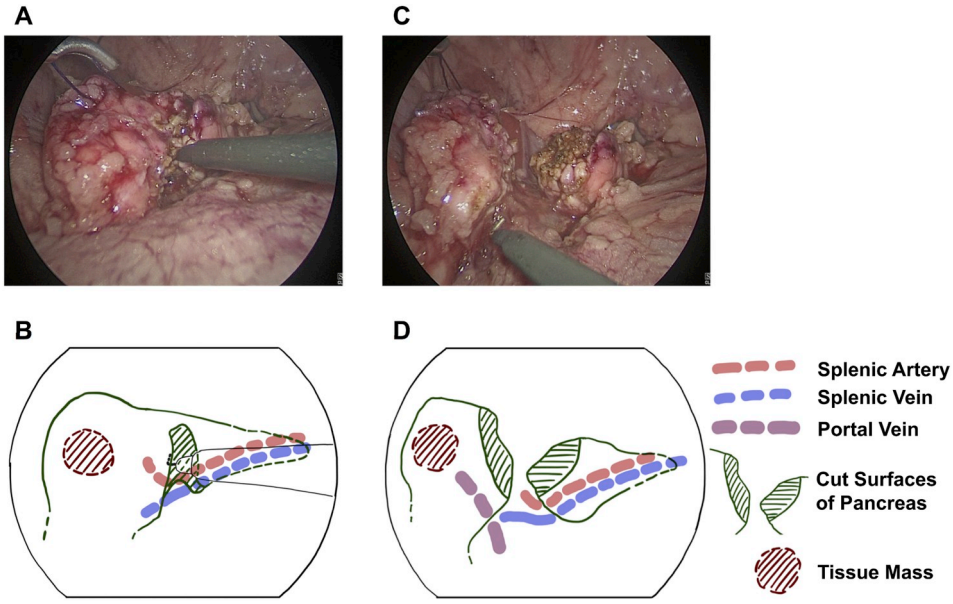

Fig. 2. A) View of laparoscopic separation of the pancreas from the splenic vessels using hook cautery. A stay stitch adjacent to the tissue mass was placed to facilitate elevation of the pancreas.
B) Schematic drawing of A).
C) Laparoscopic view after complete transection of the pancreas distal to the mass.
D) Schematic drawing of C).

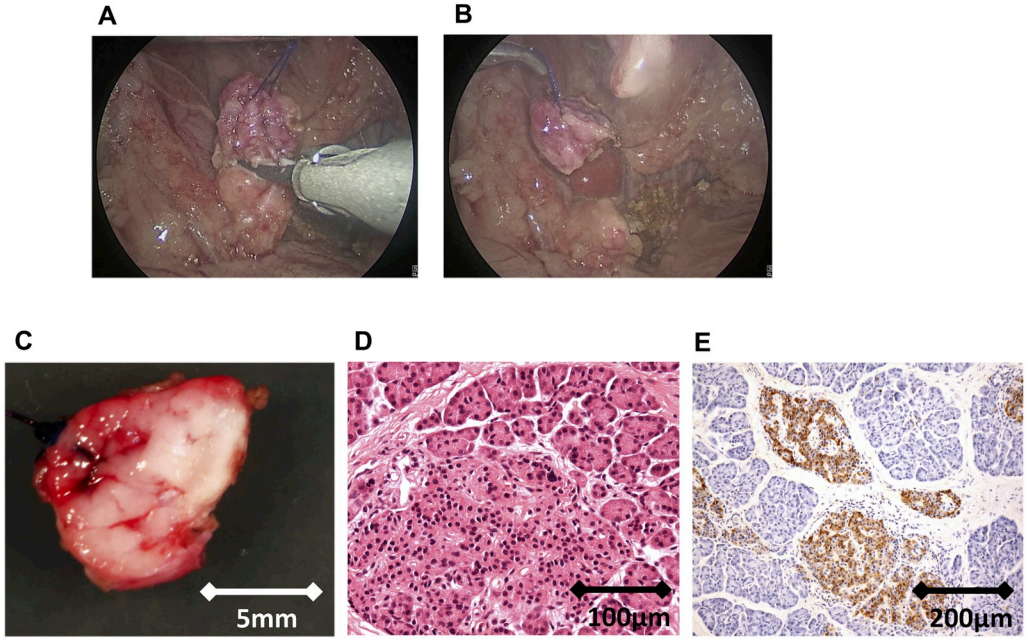

Fig. 3. A) Transection of the pancreas proximal to the mass using a Harmonic® laparoscopic scalpel, which seals the divided pancreatic duct.
B) Resected middle segment of the pancreas containing the focal lesion.
C) Macroscopic image of the removed and transected mass with the fibrous white appearance adjacent to normal pancreatic tissue.
D) Hematoxylin-Eosin stain from a paraffin section through the mass, showing islet cells with an enlarged cell nucleus as sole abnormality.
E) Immunohistochemistry stain for pro-insulin; the section is through the same area as B) and shows an increase in strongly positive endocrine tissue.

of diabetes. Second, to do an open pancreatico-jejunostomy to collect and drain the secretions produced by the pancreatic tail. Third, to perform the latter procedure laparoscopically, which we decided to do to avoid a laparotomy with all its consequences. A drainage technique for middle segmental pancreatic resections has been described after resection of benign pancreatic body lesions operated in an open fashion in adults [23,24]. An earlier case report of laparoscopic resection of a pancreatic cystadenoma in a 55 year old woman described an end-to-side anastomosis to assure drainage of the remaining distal pancreas [25].

The fact that frozen sections of a macroscopically identified mass had a normal appearance after HE-staining led to an intraoperative decision-making dilemma. We considered cautery artefact as a potential reason why sections of the mass did not display HI typical features; however, no heat-induced changes were detected in the sections through the mass. To reduce cautery artefacts, some recommend sharp dissection for enucleation and transection of the distal pancreas in case of a proximal pancreatectomy [10]. Also, focal lesions might feature tentacles extending into otherwise healthy surrounding pancreatic tissue. Therefore, confirmation of complete excision on frozen sections is generally our standard [5]. However, since not even the macroscopic mass displayed the HI-typical changes on frozen sections, we were unable to assess eventual outgrowths from the mass until we had confirmation of complete removal using immunohistochemical staining.

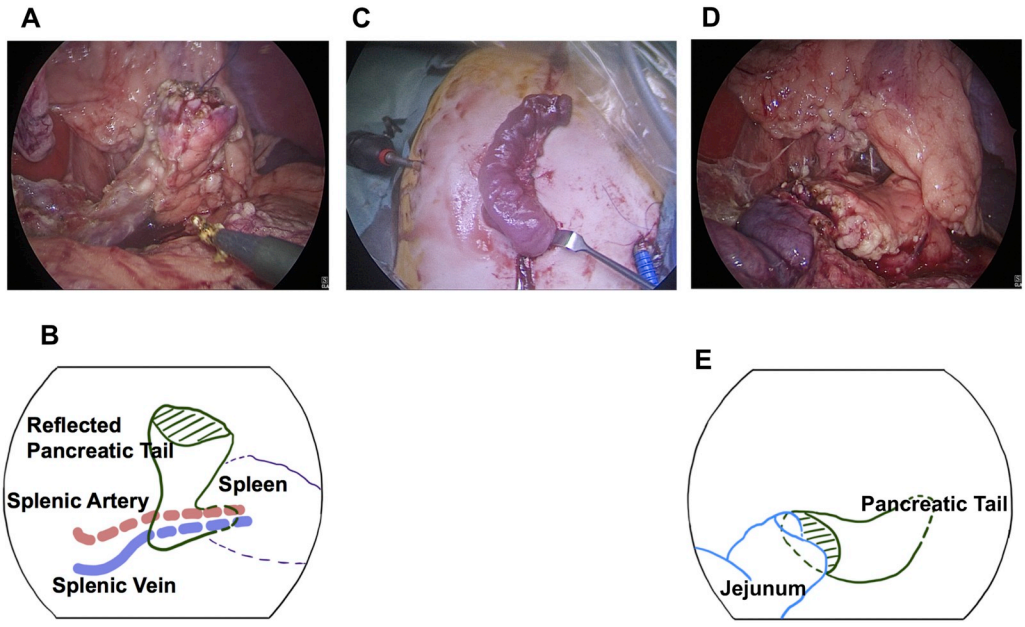

Fig. 4. A) Laparoscopic separation of pancreatic body from the underlying splenic vessels to resect a short segment of pancreas distal to the already resected neck.
B) Schematic illustration of the intraoperative situs.
C) View of the jejunal limb brought out through the slightly extended umbilical incision, prepared for the pancreatico-jejunal anastomosis after forming the foot-point anastomosis.
D) Laparoscopic view of the jejunal limb juxtaposed to the pancreatic tail after bringing the jejunum through the transverse mesocolon and fashioning the posterior part of the anastomosis.
E) Schematic representation of intraoperative situs after completion of the dorsal part of the anastomosis.

We are currently investigating whether long-standing Sirolimus treatment affects microscopic appearance of hyper productive pancreatic islet cells.

To the best of our knowledge, we are herein reporting the first paediatric case of a laparoscopically performed middle segmental pancreatic resection with laparoscopic Roux-en-Y pancreatico-jejunostomy. We suggest considering this approach for similar cases, since it combines superior intraoperative visualisation with reduced surgical trauma and thus diminished postoperative pain and favourable cosmetics.

Acknowledgement

Consent

The patient's family agreed to the anonymised publication of the case.

Author's contributions

MS: Drafting of manuscript, critical revision of manuscript, creating of figures.

PS: Critical revision of manuscript.

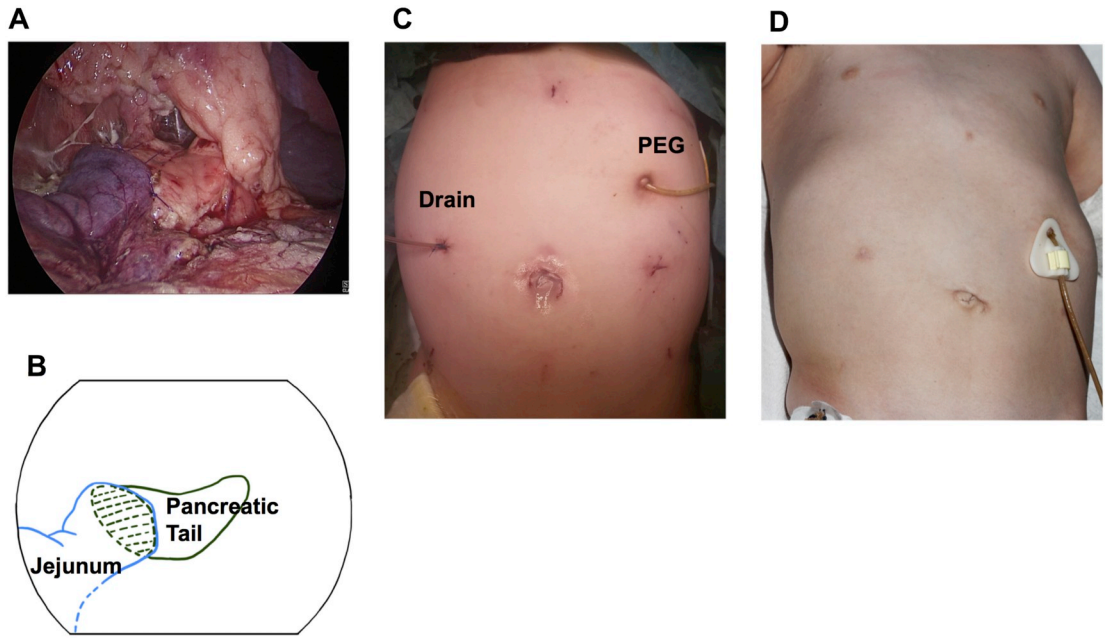

Fig. 5. A) Picture of the laparoscopically completed pancreatico-jejunal anastomosis before drain placement.
B) Schematic drawing of the pancreatico-jejunal anastomosis.
C) Photograph of the immediate postoperative appearance of the patient's abdomen with the drain and the previously placed percutaneous endoscopic gastrostomy (PEG) in situ.
D) Photograph of the patient's abdomen 3 months after the operation, showing the well healed short wounds of the trocar sites.

MA: Critical revision of manuscript.
PDC: Conception and critical revision of manuscript.

Disclosure statement

No competing financial interests exist.

Patient consent

The patient's parents agreed to the publication of the case and the included pictures.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Conflict of interest

The following authors have no financial disclosures: MS, PS, MA, PDC.

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