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CASE REPORT

Robotic-assisted, spleen-preserving distal pancreatectomy for a solid pseudopapillary tumour in a pediatric patient: a case report and review of the literature

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Abstract
Solid pseudopapillary tumour is a rare carcinoma of the pancreas with low-grade malignant potential that typically presents in females in their third decade. The tumour most commonly occurs in the tail of the pancreas, although any site can be affected. Surgical resection is the standard treatment and offers an excellent prognosis. Robotic-assisted laparoscopic surgery is an emerging technique for the treatment of pancreatic neoplasms. We report a case of a 17-year-old female with acute onset abdominal pain who was found to have a mass in the distal pancreas. A robotic-assisted laparoscopic distal pancreatectomy with spleen preservation was successfully performed, its first reported use in a pediatric patient. The patient had an uncomplicated recovery. The robotic approach can be considered for younger patients presenting with a solid pseudopapillary tumour in the distal pancreas.

Keywords Pseudopapillary · Robotic · Distal pancreatectomy · Pediatric · Pancreas

Introduction

Solid pseudopapillary tumour (SPT) of the pancreas is a rare carcinoma estimated to comprise less than 3% of exocrine pancreatic tumours, although with increased recognition the incidence has been estimated to be up to 6% [1]. Grossly, most tumours have areas of necrosis and haemorrhage resulting in both cystic and solid portions [2]. The tumours are most often well circumscribed and located in the tail of the pancreas followed by the head, although an estimated 10–15% of cases are malignant, often involving the liver or peritoneum [2, 3]. The low-grade malignant potential of the tumour leaves it amenable to surgical intervention [1, 4]. Cuschieri first reported on laparoscopic pancreatic surgery in 1994 [5]. The first case series on laparoscopic distal pancreatectomies (LDP) for SPT published by Cavallini et al. [6] in 2011 reported its safety and feasibility as a treatment for SPT, though the technical challenges of laparoscopic pancreatic surgery have resulted in its slow but rising acceptance. Robotic assistance offers several advantages as an adjunct to laparoscopic surgery, including increased dexterity and three-dimensional visualization [7]. Herein, we report the first use, to our knowledge, of robotic-assisted LDP in a pediatric patient who presented with a distal pancreatic mass.

Case report

A 17-year-old female presented to her regional hospital with a 1-week history of recurring right lower quadrant pain. The pain was of acute onset, severe but non-radiating and sharp in quality. Other symptoms included fever and several episodes of emesis. Past medical history was insignificant. Physical examination revealed a soft, non-distended abdomen with minimal tenderness in the right lower quadrant. An abdominal ultrasound was negative for appendicitis, while a subsequent computed tomogram (CT) of the abdomen revealed an incidental well-circumscribed, heterogeneous

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and hypoechoic lesion measuring 3 cm in diameter in the tail of the pancreas, consistent with a diagnosis of SPT (Fig. 1). A repeat abdominal ultrasound completed upon transfer to our institution showed a normal appendix and ovaries, and confirmed the presence of a mass in the pancreas. Due to the thickened terminal ileum noted on her CT as well as previous right lower quadrant pain and diarrhea, this patient was assessed by Pediatric Gastroenterology and underwent a colonoscopy, which revealed non-pathological lymphoid hyperplasia in the terminal ileum. The remainder of her preoperative workup included a normal chest X-ray and normal levels of pancreatic tumour markers (chromogranin A, CA199S and CA125). Her abdominal pain and diarrhea resolved. However, given the presumed diagnosis of solid pseudopapillary pancreatic tumour, informed consent was obtained from the patient and her parents to proceed with robotic-assisted LDP.

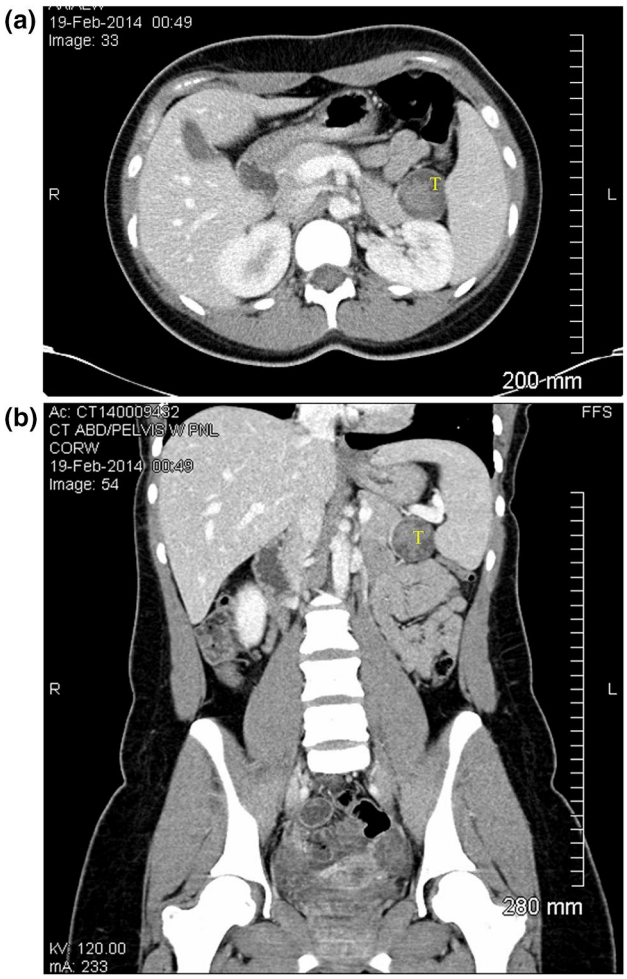

Fig. 1 Computed tomography of the abdomen reveals a well-circumscribed, heterogeneous and hypoechoic lesion in the tail of the pancreas (T)

The surgery was performed using the da Vinci Surgical System (Intuitive Surgical, Sunnyvale, CA, USA) robotic platform. The patient was positioned supine with her left flank partially elevated. Following induction of general anaesthesia and endotracheal intubation, trocar placement for the procedure was as follows: one 8 mm camera trocar infra-umbilically using the Hasson technique, one 5 mm robotic trocar in the right upper quadrant just off midline, one 5 mm robotic trocar in the left lower quadrant, one 5 mm laparoscopic trocar in the right abdomen lateral to the umbilicus and a 10/12 mm disposable trocar in the left abdomen lateral to the umbilicus (Fig. 2). The robot was then placed over the patient's left shoulder and the three robotic arms were docked to the appropriate trocars.

Subsequently, the gastrocolic omentum was incised to provide access to the lesser sac. A silk suture was used to place a hitch stitch along the posterior wall of the stomach to retract the stomach superiorly. This allowed for visualization of the pancreatic mass, which appeared non-adherent to the spleen or any other structures. Beginning superiorly and then moving inferiorly, the borders of the distal pancreas up to the mid-body were freed through blunt dissection and the splenic artery and vein were identified. The splenic artery was carefully freed and retracted using a vessel loop. A Harmonic scalpel (Ethicon, Cincinnati, OH, USA) was used to clear away fatty tissue and small vessels. A Penrose drain was placed at the mid-body to retract the pancreas and provide an adequate resection margin from the tumour. An Echelon 45 mm stapler (Ethicon, Cincinnati, OH, USA) with blue (3.5 mm) reload was used to transect the body of the pancreas. A 10 mm EndoCatch (Medtronic, Minneapolis,

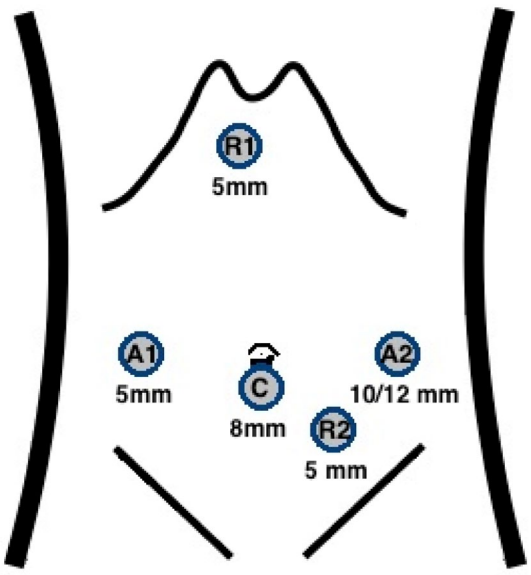

Fig. 2 Placement of trocars. The ports for the camera (C), robotic arms (R1 and R2) and the assistant ports (A1 and A2) are shown

MN, USA) bag was placed through the left 10/12 mm port to remove the distal pancreas and mass. Ensuring good hemostasis, the hitch stich was removed and we proceeded to close all five incisions. Mastisol (EloQuest Healthcare, Ferndale, MI, USA), Steri-Strips (Nexcare, Brighton, MI, USA) and Op-Site spray (Smith and Nephew, London, UK) were applied. Intraoperative blood loss was nil and operating time was 277 min.

Histological examination of the mass confirmed the diagnosis of SPT (Fig. 3). Post-operatively, the patient was stable and recovery was uneventful. Patient was discharged on post-operative day 6. Magnetic resonance imaging (MRI) done at 6 months and at 1 year post surgery showed no evidence of disease recurrence.

Discussion

Malignant pancreatic tumours occur very infrequently in the pediatric population, with an estimated age-adjusted incidence of 0.191 per million in the 0–19-year-old age group [8]. Primary tumours may be of epithelial or non-epithelial origin, the latter being very rare. Pancreatoblastoma and SPT are the most common epithelial tumours in pediatric patients, while ductal adenocarcinoma, acinar cell carcinoma and pancreatic endocrine neoplasms also occur [9]. Shorter et al. [9] from Memorial Sloan-Kettering published the most recent and largest pediatric case series of malignant pancreatic tumours in patients less than 21 years of age, and found 5 out of 17 had SPT over a 35-year period. Other smaller series from large pediatric hospitals only found 1 SPT each over a 20–30 year period [10–12]. The most frequent presenting features of pancreatic tumours are abdominal pain or an upper abdominal mass; jaundice is much less common than in adults [11]. However, over 15% of children with SPT are asymptomatic [1]. The histogenesis of SPT is unclear but they have a strong predilection for young women, in whom approximately 90% of tumours occur, with a mean age of presentation between 20 and 30 years of age [1, 2]. Although SPT are typically large (average diameter of 8–10 cm), there is normally no evidence of pancreatic dysfunction or elevated tumour marker levels [13]. The differential diagnosis includes pseudocysts, other cystic neoplasms and secondary tumours of the pancreas, with an abdominal CT scan or MRI being most useful in identifying SPT [13].

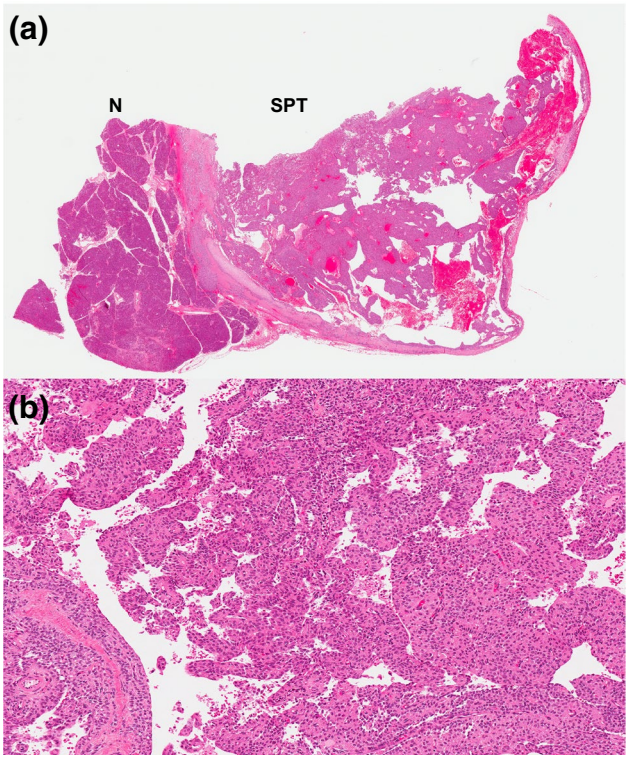

Fig. 3 Histopathology identifying **a** normal pancreatic tissue on left and encapsulated solid pseudopapillary tumour on right (H&E ×0.5) and **b** formation of pseudopapillae (H&E ×10)

Surgical resection is the standard of care for SPT. Complete resection of the tumour allows a 5-year survival rate of over 95% [1, 4]. As over 40% of tumours involve the tail of the pancreas, LDP is the most commonly used minimally invasive surgical technique. Although effective, the literature reporting this technique is restricted to a small number of case reports and case series [2, 6, 14, 15]. The close relationship of the pancreas to surrounding vessels and structures and its retroperitoneal location create a challenge for laparoscopic surgery, such that only a handful of laparoscopic pancreatic resection case series have been published [7]. However, LDP has been gaining in popularity amongst surgeons as it does not require reconstruction, thereby reducing its difficulty.

The technical challenges of LDP have resulted in laparotomy remaining the most commonly used method for SPT resection. A recent meta-analysis by Nigri et al. [16] comparing open distal pancreatectomy to minimally invasive distal pancreatectomy for a variety of benign and malignant lesions highlighted the benefits of LDP. Nigri showed that although mortality and re-operative rates did not differ between the two methods, the minimally invasive approach had fewer complications, less blood loss, a shorter time to oral intake and a shorter post-operative hospital stay.

Robotic technology provides further benefits when used as an adjunct to laparoscopic surgery, including tremor stabilization, three-dimensional visualization and wrist-like movement of instruments. A retrospective analysis of 77 patients who underwent open, laparoscopic and robotic-assisted distal pancreatectomies done by Waters et al. [17] found that while the robotic approach increased operating time by a mean of 74 min, it had a significantly higher spleen preservation rate of 65%, compared to 31% in LDP.

Another retrospective comparison of 45 patients done by Kang et al. [18] also found a significantly increased splenic preservation rate with the robotic approach (95 vs. 64% with LDP). The value of splenic preservation in distal pancreatectomy has been previously reported by Shoup et al. [19], who found that perioperative infections, blood loss and length of hospital stay were significantly increased following splenectomy. Post-operatively, preserving the immunological role of the spleen reduces the risk of infection and sepsis [20]. Another advantage of the robotic approach is a reduced rate of conversion to laparotomy, with Daoudi et al. [21] reporting a rate of 0 vs. 16% in LDP. In our case, we believe that the higher magnification and three-dimensional visualization of the robotic camera as well as the wrist-like instrumentation of the robotic instruments were key elements in enabling us to carefully dissect the splenic vessels off the distal pancreas and preserve the spleen. We agree with Goh et al. [22] that an additional expert surgeon at the bedside is important for suctioning and retraction, as well as assistance with rapid conversion to laparotomy if a complication arises. Overall, we found the additional operative time incurred with the use of the robot is offset by the numerous advantages listed.

Conclusion

We report a case of a 17-year-old female with a SPT in the tail of the pancreas that was successfully treated with spleen preserving robotic-assisted LDP. We found that the advantages provided by a robotic approach, including improved surgical dexterity and visualization, allow for its use as a safe and effective alternative to open pancreatectomy or LDP in the pediatric patient.

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Compliance with ethical standards

Conflict of interest Mr. Raj Lalli and Drs. Neil Merritt, Christopher M. Schlachta and Andreana Bütter declare that they have no conflict of interest.

Ethical standards All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors.

Consent Written informed consent was obtained from the patient for publication of this Case Report/any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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