Challenging the limits in pancreatic surgery: A case report

Johannes Lemke a, Stefan A. Schmidt b, Marko Kornmann a, Karl-Heinz Orend c, Doris Henne-Bruns a,∗

a Department of General and Visceral Surgery, University of Ulm, Albert-Einstein-Allee 23, 89081 Ulm, Germany
b Department of Diagnostic and Interventional Radiology, University of Ulm, Albert-Einstein-Allee 23, 89081 Ulm, Germany
c Department of Cardiothoracic and Vascular Surgery, University of Ulm, Albert-Einstein-Allee 23, 89081 Ulm, Germany

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A B S T R A C T
INTRODUCTION: Today, pancreatic surgery can be performed with low mortality and tolerable morbidity in specialized centers. Nevertheless, due to its anatomical localization and proximity to important vascular structures, surgical resection of the pancreas remains challenging in many cases.

PRESENTATION OF CASE: Here, we present the case of a young woman who presented in our department with abdominal pain and a tumor mass located at the pancreatic head. She had undergone explorative laparotomy elsewhere before, in which the pancreatic tumor mass was reported to be unresectable due to infiltration of the mesenteric root. However, biopsies obtained had not revealed malignancy. Moreover, postoperatively a stenting of the portal vein had been performed due to portal vein thrombosis and varices. Upon admission in our clinic, computed tomography revealed a tumor of the pancreatic head, occlusion of the portal vein stent and, more importantly, extravascular dislocation of the stent with perforation into the stomach. Upon explorative laparotomy we initially performed a mesenterico-caval shunt to release portal hypertension. Secondly, the dislocated stent was successfully removed after gastrotomy, and finally, a partial pancreatocoduodenectomy was performed. Interestingly, the histopathological analysis revealed granulocytic epithelial lesions (GELs) confirming a type-2 autoimmune pancreatitis without evidence for malignancy. The postoperative course was uneventful and the patients was dismissed without any remaining symptoms.

DISCUSSION & CONCLUSION: This interesting and unique case underlines the complexity in diagnosis of pancreatic tumors with unresolved dignity and differential diagnosis of pancreaticitis and pancreatic cancer. Furthermore, it demonstrated the challenges in pancreatic surgery for exceptional and uncommon conditions demanding complex surgical approaches.

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1. Introduction
Pancreatic tumors comprise benign lesions and malignant neoplasms, most importantly adenocarcinomas arising from the pancreatic ducts [1]. Pancreatic ductal adenocarcinomas have a dismal prognosis with a 5-year survival rate of about 5% [2]. The only chance for cure is provided by surgical resection which is the therapy of choice for suspected, resectable pancreatic cancer [3]. Despite major improvement in radiological diagnostic, preoperative differentiation between pancreatic cancer and benign lesions including chronic pancreatitis remains imperfect in many cases [4]. The area of major pancreatic surgery arose at the turn of the 19th and 20th century when the first distal pancreatectomy and first partial pancreatocoduodenectomy had been successfully performed [5]. However, until the 1980s pancreatic resection was still associated with perioperative mortality up to 30% and significant morbidity [5]. Therefore, the actual benefit of major pancreatic surgical approaches had been a matter of debate for long time. Due to establishment of high volume centers with experienced pancreatic surgeons and major advances in intensive care treatment mortality has dropped below 5% since the 1980s [6]. Consequently, today surgery provides a safe therapeutic approach for pancreatic conditions, most importantly for pancreatic tumor of unsolved or confirmed malignant dignity [7]. However, due to its anatomical proximity to main vascular structures such as coeliac trunk, superior mesenteric artery and mesenteric/portal vein, pancreatic resection remains a major challenge in surgery. In particular this holds true in cases that require complex vascular re-construction upon resection, which usually also determine limitations in pancreatic surgery [8]. Here, we present a unique cases in which partial pancreatectomy in combination with mesenterico-caval shunt was required for an unclear mass of the pancreatic head with portal-caval collateralization and extravascular dislocation of a previously implanted portal vein stent.
2. Presentation of case

We report on a 19-year-old woman who had been primarily admitted to the department of internal medicine in April 2011 with the history of pancreatitis of unclear nature 4 months before. The patient reported an uneventful medical history without preexisting conditions or traumas in the past and there was no family history of autoimmune diseases. The patient was not under any long-term medication. Moreover, there was no evidence for alcohol or drug abuses. At that time a computed tomography had revealed a portal vein thrombosis as well as a tumor mass located at the pancreatic head suspicious for pancreatic cancer. Subsequently, the patient was referred to an external surgical center where an explorative laparotomy was performed. Intraoperatively, the tumor mass was regarded to be unresectable due to tumor infiltration of the mesenteric root and, subsequently, multiple biopsies of the tumor mass were obtained. Postoperatively, a portal venous stent was placed interventionally due to portal vein thrombosis and varices. Interestingly, at that time the pancreatic biopsies did not reveal any evidence for malignancy and the patient was dismissed with the diagnosis of a chronic pancreatitis of unknown specificity.

In March 2015 the patient reported on abdominal pain. Imaging confirmed the diffuse and large tumor mass located at the pancreatic head being constant in size compared to previous CT scans with intra- and extrahepatic cholestasis. However, in addition, occlusion of the portal vein stent was detected with pronounced portal-systemic collateralization and varices. Intriguingly, the previously implanted portal vein stent was found to be dislocated with perforation into the stomach (Fig. 1). Carbohydrate-Antigen 19.9 (CA19.9) was slightly elevated at that time. Subsequently, the patient was admitted to our surgical clinic for further treatment. In summary, we were confronted with (i) a pancreatic tumor with still unsolved dignity with compression of the duodenum and cholestasis (ii) occlusion of the previously implanted portal vein stent with portal-systemic collateralization and varices, and (iii) extravascular dislocation of a portal vein stent with penetration into the stomach. Although, retrospectively, the existence of a pancreatic adenocarcinoma already a time of first explorative laparotomy in 2011 was highly unlikely, the progression to a malignant tumor on the basis of chronic pancreatitis had to be considered, in particular due to increasing symptoms and cholestasis. Moreover, the dislocated portal venous stent was associated with a high risk of contamination with sepsis as well as acute hemorrhage. Moreover, interventional release of cholestasis by endoscopic retrograde cholangiography (ERC) appeared unfeasible due to duodenal compression. Upon discussion this complex case in an interdisciplinary meeting we decided on a surgical approach with the aim to (i) ultimately confirm dignity of the pancreatic tumor mass, (ii) release portal hypertension to alleviate esophageal varices, (iii) ensure biliary drainage and gastro-intestinal passage, and (iv) allow for recovery of the dislocated stent. Upon explorative laparotomy we firstly exposed the portal vein and the mesenteric root by carefully transecting all varices. In the next step a mesenterico-caval shunt to release the portal hypertension and congestion of the small intestine was performed by direct mesenterico-caval anastomosis after careful dissection and mobilization of all structures of the mesenteric root. Having successfully accomplished this, we dissected the hepatoduodenal ligament to identify the portal vein, hepatic artery and central bile duct. Subsequently the dislocated and stomach-penetrating stent was removed (Fig. 2). The portal vein was ligated because of the minimal residual flow in the mainly occluded vessel. Finally, a partial duodenopancreatectomy and resection of the distal stomach (including the perforation site) with hepatocyojejunostomy and gastro-jejunostomy were performed to achieve ultimate cure for the unclear pancreatic mass. Due to fibrotic alteration of the pancreatic tissue the pancreatic duct could not be identified and the pancreatic stump oversewn with non-absorbable sutures was blindly closed by stiches. Postoperatively, the patient was monitored on our intensive care unit. The further clinical course was uneventful. Pathological analysis revealed a pancreatitis with acinar atrophy, ductal ectasia and inflammatory exudate without evidence for malignancy. Intriguingly, further histopathological analysis detected granulocytic epithelial lesion (GELs), a hallmark for a type-2 autoimmune pancreatitis (AIP). The patient is currently followed-up in our department. Postoperatively all symptoms vanished and the patient enjoys a good quality of life.

3. Discussion

This interesting and very unique case report demonstrates, on the one hand, the complexity and complicacy in the diagnosis of (autoimmune) pancreatitis compared to benign and malignant tumors of the pancreas. On the other hand, it highlights the technically challenges (and possibly limits) which may be encountered in pancreatic surgery. AIP is a very rare and benign inflamma-
Intraoperative images show the removed stent penetrating the stomach (white arrow). (B) White arrow indicates the mesenteric-caval shunt.

Fig. 2. (A) Intraoperative images show the removed stent penetrating the stomach (white arrow). (B) White arrow indicates the mesenteric-caval shunt.

tory condition of the pancreas and comprises two subtypes, type-1 and type-2 AIP [9]. Both types have in common they are associated with immune cell infiltration in the pancreas leading to an auto-inflammatory response. Although both subtypes are considered to differ in epidemiological, immunological and pathological features such as the type of immune cell infiltration, both have in common that patients commonly respond to steroid treatment. This matter of fact may be also helpful as a diagnostic tool to distinguish AIP from pancreatic of other origin [10]. Preoperatively a steroid treatment was initiated in our patient which led to some clinical response in terms of decreasing cholestasis which supported the diagnosis of AIP. Postoperatively, the histopathological detection of GEL in the specimen upon resection confirmed the diagnosis von type-2 AIP. However, preoperatively, a pancreatic malignancy could not be definitely excluded in the performed diagnostic. Moreover, a primary surgical approach was pursued in our case because of the portal vein thrombosis with progressive portal-caval transformation and varices, and in addition, a dislocated and stomach-penetrating portal vein stent. Upon explorative laparotomy we performed a combination of two itself unquestionable complex surgical procedures: a mesenterico-caval shunt and a partial duodenopancreatectomy. This approach was further complicated by pronounced cavernous transformation and varices as well as a dislocation of a stomach infiltrating stent which had to be recovered.

4. Conclusion

To our knowledge this case report is the first description of this extremely challenging combination of above described surgical procedures. In conclusion, this report highlights challenges in pancreatic surgery which, however, may be successfully overcome by complex surgical strategies reported here.

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. This case report is in line with the SCARAE criteria [11]. All author declare no conflict of interest.

Conflicts of interest

All authors declare no conflict of interest.

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Ethical approval

Not applicable.

Consent

Written consent of the patient has been obtained.

Author contribution

JL, MK, KHO and DHB designed the study, JL and DHB performed the research and wrote the manuscript. S.A.S. provided radiological analysis. All authors read and approved the manuscript.

Registration of research studies

Not applicable.

Guarantor

Prof. Dr. Doris Henne-Bruns.

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