Successful outcomes after laparoscopic spleen-preserving pancreatic resection for a desmoid tumor: A case report

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1. Introduction

Desmoid tumors are rare benign neoplasms that result from the abnormal proliferation of fibroblasts. These tumors are locally aggressive but do not metastasize [1]. The reported incidence ranges from 2.4 to 4.3 per 1,000,000 individuals/year [2]. Isolated pancreatic desmoid tumors are extremely rare. Among the 27 cases of pancreatic desmoid tumors reported in the English literature to date [3], only one patient has been treated with laparoscopic resection [4]. Therefore, the use of this technique to treat pancreatic desmoid tumors has not been verified in literature. Laparoscopic distal pancreatectomy is associated with faster recovery and less morbidity than open surgery [5]. We present a rare case of a pancreatic desmoid tumor in a patient who was successfully treated with laparoscopic spleen-preserving pancreatic resection. This work has been reported in line with the Surgical Case REport (SCARE) guidelines [6].

2. Presentation of case

A 60-year-old male patient was referred to our hospital for evaluation of back pain. His past medical and family history was unremarkable. Abdominal ultrasound showed a hypoechoic mass with clear margins measuring 30 mm in diameter in the pancreatic tail. A contrast-enhanced computed tomography (CT) scan showed a mass protruding from the pancreatic tail with no dilatation of the main pancreatic duct (Fig. 1). The mass was circumscribed and there was delayed contrast enhancement. Abdominal magnetic resonance imaging was performed for further characterization of the lesion; the mass presented as low intensity on T1-weighted imaging and slightly high intensity on T2-weighted imaging (Fig. 2). On the basis of the above findings, we initially suspected a solid pseudopapillary tumor or a gastrointestinal stromal tumor. The patient underwent laparoscopic evaluation which revealed that the edge of the tumor was relatively distinct, but strongly adherent to the pancreas (Fig. 3). Subsequently, laparoscopic spleen-preserving distal pancreatectomy was performed for radical excision. The operating time was 255 min, and blood loss was minimal. The patient did not have any complications and was discharged 10 days after surgery.

Abbreviations: CT, computed tomography; FAP, familial adenomatous polyposis.
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connective tissue (Fig. 4). Pathological analysis confirmed that the resection was complete. Immunostaining was negative for s-100, desmin, and c-kit, and positive for β-catenin and actin-smooth muscle. The final diagnosis was of a desmoid tumor embedded within the pancreatic tail. There was no evidence of recurrence at 36 months of follow-up in clinical examinations including dynamic CT.

3. Discussion and conclusion

A desmoid tumor is a fibrous soft tissue tumor arising in the fascia and musculoaponeurotic tissues. Stout et al. [7] demonstrated that desmoid tumors are characterized by abnormal proliferation of fibroblasts, infiltrative growth pattern, lack of malignant findings, absence of metastases, presence of collagen fibers in intercellular space, and local recurrence. Desmoid tumors can be found in any of the fibrous connective tissues throughout the body [8]. The etiology of these tumors is unknown, but they tend to occur sporadically or in association with familial adenomatous polyposis (FAP) [8]. Reported risk factors of desmoid tumors are a history of trauma, positive family history, pregnancy, surgery, use of contraceptives, genetic mutation, irradiation, and FAP [2]. In our case, there was no personal or family history of FAP, and the patient did not have any of the other above-mentioned risk factors. Thus, this was a sporadic case.

Pancreatic desmoid tumors are extremely rare, with only 27 cases reported in the English literature to date [3]. Gerleman et al. [1] reviewed 17 cases of pancreatic desmoid tumors and found that the majority (58%) of tumors involved the pancreatic tail, and only 3 cases were associated with a genetic disorder such as FAP or with systemic disease. In the present case, the patient was diagnosed with a sporadic isolated pancreatic desmoid tumor strongly involving the pancreatic tail.

In general, preoperative diagnosis of desmoid tumors is difficult, as these tumors do not have distinct clinicopathologic features [3]. Desmoid tumors tend to present as solid and homogeneously dense masses on CT scan, but the degree of enhancement varies among cases. In our case, CT scan showed a homogeneous mass with no cystic components and a delayed contrast effect. Our preoperative differentials were of a solid pseudopapillary tumor or a gastrointestinal stromal tumor. The final diagnosis of a desmoid tumor requires histological and immunohistochemical analysis. Although a desmoid tumor appears well defined at gross analysis, at the microscopic level its margins appear to infiltrate the adjacent structures [4]. In our case, there was a significant invasion of the tumor into the pancreatic parenchyma. Immunohistochemically, the tumor cells are negative for CD34, CD117, and S-100 protein [8]. These findings exclude gastrointestinal stromal tumors and neurogenic tumors. Nuclear staining for β-catenin is a consistent finding in more than 80% of cases [3], and these features allowed us to confirm the diagnosis of a desmoid tumor.

Surgery is the first-line treatment for resectable desmoid tumors [2]. Achieving clear margins is essential because surgical margin status correlates with the risk for recurrence [9]. In our case, as the tumor did not infiltrate the extrapancreatic splenic vein and splenic artery, resection of the pancreatic tail with laparoscopic spleen-preserving surgery was performed.
To the best of our knowledge, this is the first case of successful treatment of a pancreatic desmoid tumor with laparoscopic surgery. Recently, laparoscopic pancreatic surgery has become widely accepted and preferable to open surgery because of its minimally invasive approach. Laparoscopic distal pancreatectomy is associated with less blood loss and fewer overall complications than open distal pancreatectomy [5]. The most common indications for laparoscopic pancreatic resection are benign or low-grade malignant tumors. We performed laparoscopic spleen-preserving pancreatic resection to minimize the risk of post-splenectomy sequelae including sepsis, thrombocytosis, thrombosis, and cancer [10]. However, laparoscopic resection of desmoid tumors should be carefully considered given the high local recurrence rate of these tumors. Laparoscopic pancreatic enucleation may be considered in certain cases, but this procedure is associated with an increased risk of positive margin status. Therefore, laparoscopic spleen-preserving pancreatectomy was considered to be the optimal surgical procedure for resection of the desmoid tumor in our patient. Another advantage of laparoscopic surgery to open surgery is that this procedure has a reduced risk of desmoid recurrence because of less scar formation. Adjuvant therapies such as systemic chemotherapy and radiotherapy have also been used for patients with desmoid tumors who are considered to have high surgical risk [3]. However, there are currently no universal clinical guidelines for the management of desmoid tumors. The patient described in this case did not receive any adjuvant therapy.

The recurrence rate of desmoid tumors is 22–85% and the mortality rate is 10–60% [2]. Our patient is currently disease-free 36 months after surgery, and remains under regular follow-up because of the high risk of tumor recurrence.

In summary, we report a rare case of a patient with a pancreatic desmoid tumor that was successfully treated with laparoscopic resection. Laparoscopic surgery appears to be a safe and effective procedure for desmoid tumors.

Conflicts of interest

None

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Ethical approval

The Institutional Review Board of Kawasaki Municipal Kawasaki Hospital approved this study and the submission (approval serial number: 201933).

Consent

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.

Author contribution

KH: Conceptualization, investigation, and writing—review and editing. KM: Writing—original draft, review, and editing of the manuscript. KA: Resources and writing—review and editing of the manuscript. SM: Writing—review and editing of the manuscript and supervision.

Registration of research studies

N/A.

Guarantor

KM and SM accept full responsibility for the study and guarantee its accuracy.

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