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Contents

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MINIREVIEWS

1242 Radiation therapy for extrahepatic bile duct cancer: Current evidences and future perspectives
Koo T, Park HJ, Kim K

1253 Antibiotics and immunotherapy in gastrointestinal tumors: Friend or foe?
Yan C, Tu XX, Wu W, Tong Z, Liu LL, Zheng Y, Jiang WQ, Zhao P, Fang WJ, Zhang HY

ORIGINAL ARTICLE

Basic Study

1262 Elevated levels of interleukin-1β, interleukin-6, tumor necrosis factor-α and vascular endothelial growth factor in patients with knee articular cartilage injury
Wang ZW, Chen L, Hao XR, Qu ZA, Huang SB, Ma XJ, Wang JC, Wang WM

Retrospective Cohort Study

1270 Anti-hepatitis C virus therapy in chronic kidney disease patients improves long-term renal and patient survivals
Chen YC, Li CY, Tsai SJ, Chen YC

Observational Study

1282 Clinical features of syphilitic myelitis with longitudinally extensive myelopathy on spinal magnetic resonance imaging
Yuan JL, Wang WX, Hu WL

Prospective Study

1291 Application of pulse index continuous cardiac output system in elderly patients with acute myocardial infarction complicated by cardiogenic shock: A prospective randomized study
Zhang YB, Zhang ZZ, Li JX, Wang YH, Zhang WL, Tian XL, Han YF, Yang M, Liu Y

META-ANALYSIS

1302 Efficacy and safety of tranexamic acid in elderly patients with intertrochanteric fracture: An updated meta-analysis
Zhou XD, Li J, Fan GM, Huang Y, Xu NW

CASE REPORT

1315 Lupus enteritis as the only active manifestation of systemic lupus erythematosus: A case report
Gonzalez A, Wadhwa V, Salomon F, Kaur J, Castro FJ

1323 Development of a biliary multi-hole self-expandable metallic stent for bile tract diseases: A case report
Kobayashi M

1330 Paraneoplastic leukemoid reaction in a patient with sarcomatoid hepatocellular carcinoma: A case report
Hu B, Sang XT, Yang XB

1337 Multiple synchronous anorectal melanomas with different colors: A case report
Cai YT, Cao LC, Zhu CF, Zhao F, Tian BX, Guo SY

1344 Huge primary dedifferentiated pancreatic liposarcoma mimicking carcinosarcoma in a young female: A case report
Liu Z, Fan WF, Li GC, Long J, Xu YH, Ma G

1351 A large basal cell adenoma extending to the ipsilateral skull base and mastoid in the right parotid gland: A case report
Du LY, Weng XH, Shen ZY, Cheng B

1358 Novel *ATL1* mutation in a Chinese family with hereditary spastic paraplegia: A case report and review of literature
Xiao XW, Du J, Jiao B, Liao XX, Zhou L, Liu XX, Yuan ZH, Guo LN, Wang X, Shen L, Lin ZY

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Contents

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CASE REPORT

Huge primary dedifferentiated pancreatic liposarcoma mimicking carcinosarcoma in a young female: A case report

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Abstract

BACKGROUND

Pancreatic liposarcoma is a rare tumor. According to a literature review, the patient described in this study is the seventh case of pancreatic liposarcoma reported in the English literature and the third case of dedifferentiated liposarcoma. Furthermore, this case had the largest primary tumor volume, and a primary pancreatic liposarcoma was diagnosed based on sufficient evidence.

CASE SUMMARY

We here report a rare case of a 28-year-old female with a huge dedifferentiated liposarcoma in the pancreatic tail. In June 2015, the patient underwent distal pancreatectomy with splenectomy. During the operation, a huge liposarcoma of approximately 28.0 cm × 19.0 cm × 8.0 cm was found, which had a yellow and white fish-like incisal surface. Based on both pathology and *MDM2* gene amplification, the tumor was diagnosed as a dedifferentiated liposarcoma. The patient was treated with surgery but declined postoperative chemotherapy. She was well at the 26-mo follow-up, and no relapse was observed.

CONCLUSION

Pancreatic liposarcoma has a low incidence. Chemotherapy should be included in the treatment regimens. Complete resection is the only effective treatment.

Key words: Pancreatic liposarcoma; Huge tumor; Distal pancreatectomy and splenectomy; Chemotherapy; Case report

Core tip: Pancreatic liposarcoma is a rare tumor. We report a case of a 28-year-old female with a huge dedifferentiated liposarcoma in the pancreatic tail, 28.0 cm × 19.0 cm

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× 8.0 cm in size, with a yellow and white fish-like incisal surface. According to a literature review, this is the seventh case of pancreatic liposarcoma reported in the English literature and the third case of dedifferentiated liposarcoma. Furthermore, this case had the largest primary tumor volume, and a primary pancreatic liposarcoma was diagnosed based on sufficient evidence. The patient was treated with surgery but declined postoperative chemotherapy. She was well at the 26-mo follow-up, without relapse.

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INTRODUCTION

Based on molecular morphology, liposarcoma can be divided into well-differentiated/dedifferentiated liposarcoma, myxoid/round cell liposarcoma, and pleomorphic liposarcoma[1-3]. For dedifferentiated liposarcoma, it is thought that a malignant non-fat sarcomatous region exists in the well-differentiated liposarcoma area at the same time. To date, six cases of pancreatic liposarcoma have been reported in the English literature[4-9]. In the present study, a patient with a huge dedifferentiated liposarcoma in the pancreatic tail is reported. Pathology after operation revealed coexisting areas of well-differentiated liposarcoma, dedifferentiated liposarcoma, and the tissue of pancreatic canal. *MDM2* gene amplification by fluorescence *in situ* hybridization showed a dedifferentiated liposarcoma that did not originate from the retroperitoneum.

CASE PRESENTATION

Chief complaints

A 28-year-old female presented with left upper abdominal discomfort with nausea and vomiting for more than 2 mo and fever for 2 wk.

History of present illness

The patient developed left epigastric discomfort with postprandial nausea and vomiting 2 mo previously without obvious predilection, the vomit consisted of stomach contents and abdominal distension was relieved after vomiting. She attended a local hospital for gastroscopy and was diagnosed with chronic atrophic gastritis, but no significant improvement was achieved after symptomatic treatment including acid inhibition and gastric preservation. She also had fever and chills for 2 wk, and as her temperature reached 39 °C, she was sent to the People's Liberation Army 202 Hospital, where an abdominal mass was found during computed tomography (CT) examination. The patient was then transferred to our hospital for further diagnosis and treatment. She was mentally stable, with a poor diet and poor sleep, with no obvious defecation and urinary abnormalities, and her weight loss was approximately 8 kg.

History of past illness

None.

Personal and family history

None.

Physical examination upon admission

The sclera showed no yellow staining, and no bleeding spots or petechiae were observed in the skin mucosa. No liver palm or spider mole was present, superficial lymph nodes were not palpable, the left epigastrium was slightly distended, gastrointestinal peristaltic wave was not observed, the abdomen was soft without tenderness, Murphy's sign was negative, and the liver and spleen were not palpable under the rib. The mass was palpable in the left epigastric region and was

approximately 15 cm in diameter. There was no muscle tension or rebound pain, no percussion pain in the liver and spleen area, shifting dullness was negative, and bowel sounds were 3-4 times/min, with no smell, air water sound, or high-pitched bowel sounds.

Laboratory examinations

Carcinoembryonic antigen was 0.76 ng/mL, alpha-fetoprotein was 2.14 ng/mL, cancer antigen 12-5 was 45.40 U/mL, carbohydrate antigen 19-9 was 6.13 U/mL, total bilirubin was 8.80 μmol/L, and direct bilirubin was 4.70 μmol/L.

Imaging examinations

The enhanced pancreatic 3D-CT images suggested a huge mass in the left upper abdomen with low density and a maximum cross-sectional area of 14.0 cm × 18.0 cm. The plain CT value was 16-24 HU.

Multiple vascular shadows were observed in the arterial phase, and the enhanced CT value was 60-95 HU in the lag period. Compressed and tortuous superior and inferior mesenteric arteries and veins were observed. A pancreatic tumor was considered, and pancreatic carcinosarcoma was not excluded (Figure 1).

Preoperatively, pancreatic carcinosarcoma was suspected, and the patient underwent surgery. Intra-operatively, the transverse colon, stomach, and jejunum were compressed by the huge tumor, which had an irregular shape, an acceptable boundary, and a clear boundary with the retroperitoneum, suggesting that the tumor was originated from the pancreatic tail. Complete resection of the pancreatic tail was performed. A mass with a proximal margin of 1.0 cm of normal pancreatic tissue was removed (Figure 2A). The inside of the mass was fish-like in texture and yellow-white. The distal pancreatic tissue was normal. Morphology inside the tumor showed no normal tissue (Figure 2B). Thus, a pancreatic source was considered and not retroperitoneal encapsulated pancreas as suggested by visual observation.

Postoperative pathology showed well-differentiated adipose tissue. Various cell sizes and shapes were observed with different trachychromatic atypia nuclei and a coexisting pancreatic canal (Figures 3A, C). Spindle cell hyperplasia was also observed when the coexisting area of undifferentiated liposarcoma and pancreatic canal were examined (Figures 3B, D). The pancreatic source was microscopically verified. Immunohistochemistry results showed the following: CK (-), vimentin (+), CD34 (+), CD117 (±), smooth muscle actin (-), S-100 (±), Dog-1 (±), CD68 (+), Desmin (-), MyoD1 (focus+), Bcl-2 (+), Beta-catenin (-), and Ki-67 (25%+). In order to verify whether the tumor was an undifferentiated liposarcoma, had a pancreatic source, and local infiltration and metastasis were present, *MDM2* gene amplification of the mass and adjacent normal retroperitoneal tissue was performed. *MDM2* gene amplification of the mass was positive (Figure 4A), while that of adjacent retroperitoneal tissue was negative (Figure 4B). This reconfirmed that the mass was pancreatic in origin and was a dedifferentiated liposarcoma.

FINAL DIAGNOSIS

Primary dedifferentiated pancreatic liposarcoma.

TREATMENT

Surgery.

OUTCOME AND FOLLOW-UP

The patient declined chemotherapy, but was well at her 26-mo follow-up, without relapse.

DISCUSSION

Dedifferentiated liposarcoma is a frequent type of liposarcoma that can occur in any part of the body[10]. It mainly occurs in the head, neck, limbs, and retroperitoneum[11-14]. Studies have shown that it amplifies to the 12q13-15 area[15]. Genes such as *MDM2* and *CDK4* are included in this area, with *MDM2* the most constantly expressed gene[16-19]. Therefore, high expression of *MDM2* plays a vital role in diagnosing dedifferentiated

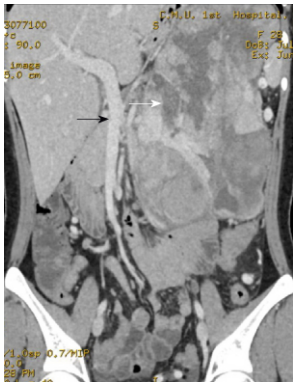

Figure 1 Preoperative computed tomography. Preoperative computed tomography shows a huge cystic-solid mass in the left upper abdomen, which compressed the superior mesenteric vein (Black arrow); necrosis can be seen in the mass (White arrow).

liposarcoma. Although liposarcoma is a common tumor, pancreatic liposarcoma is rare. To date, there are only six cases of primary pancreatic liposarcoma reported in the English literature[4-9] (Table 1). According to a review of the existing literature, it is essential to confirm whether the tumor is a differentiated liposarcoma and to assess whether it originates from the pancreas. We suggest that this tumor can be diagnosed according to the following aspects: (1) Preoperative CT revealed pancreatic occupancy with an iconographic "interspace" in the retroperitoneum, stomach, colon, and small intestine; (2) A mass located in the pancreas or a pancreatic source was found during surgery, which could be completely cut out by blunt and sharp dissection; (3) Post-operative microscopic examination detected the co-existing area of the pancreatic canal, a well-differentiated liposarcoma, and undifferentiated liposarcoma; (4) *MDM2* gene amplification by fluorescence *in situ* hybridization examination confirmed that the mass had a positive amplification, and the retroperitoneum or other control tissues had a negative amplification; and (5)There was no evidence of liposarcoma at the other sites.

CONCLUSION

Most cases are diagnosed *via* surgery combined with pathology. One of the cases in the literature was diagnosed by positive amplification of *MDM2*. However, from the comprehensive review, the diagnosis lacked evidence, which is the key in diagnosing this disease. In addition, we did not exclude the pancreatic source according to preoperative CT, and the first histological report revealed a pancreatic liposarcoma. We revised the diagnosis following *MDM2* detection and re-evaluated the microscopic hematoxylin and eosin staining. In all seven cases, it was found that the tumor may be derived from the pancreatic matrix, and evidence of a pancreatic source is not enough. This tumor has a poor prognosis when the onset site is the pancreatic tail. Therefore, post-operative chemotherapy was suggested for the patient in the present study with gemcitabine and cis-platinum, but it was declined. The patient was well at the 26-mo follow-up, without relapse. We conclude that this tumor has a low incidence. When a patient is diagnosed with liposarcoma, chemotherapy should be included in the treatment regimen. Complete resection is the only effective treatment.

Table 1 Previously reported cases of pancreatic liposarcoma

First author	Age/sex	Symptoms	Yr	Liposarcoma subtype	Liposarcoma size	Treatment	Outcome	Evidence of pancreatic origin
Elliott	59/F	Abdominal distension	1980	Pleomorphic	16 cm	DP	6 yr	Surgery
Dodo	76/M	Abdominal pain	2005	Well differentiated with area of de-differentiation	9 cm	DP	26 mo	Surgery
Kuramoto	24/M	Abdominal distension	2013	Myxoid	25 cm	MP	44 mo	Surgery
Machado	42/M	Abdominal pain	2016	Dedifferenti-ated with high grade components	6.8 cm	DP	5 yr	Intrapancreatic
Matthews	65/F	None	2016	Well differentiated	4 cm	DP	none	Intrapancreatic + adjacent retroperitoneal *MDM2* FISH
Han	29/F	None	2017	Dedifferenti-ated	20 cm	DP	1 yr	Surgery
Present case	28/F	Abdominal pain		Dedifferenti-ated with high grade components	28 cm	DP	26 mo	Surgery + histology + adjacent retroperitoneal *MDM2* FISH

F: Female; M: Male; DP: Distal pancreatectomy; CP: Central pancreatectomy; MP: Middle pancreatectomy; FISH: Fluorescence *in situ* hybridization.

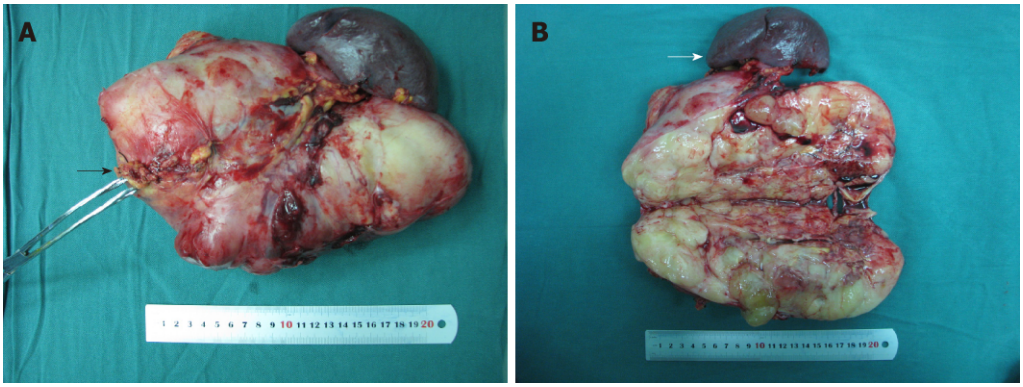

Figure 2 Surgical procedure. The pancreatic tail was removed. A: The giant mass in the pancreatic tail was approximately 28.0 cm × 19.0 cm × 8.0 cm. The black arrow shows the normal pancreatic tissue; B: The mass was longitudinally opened. The gray-white and fish-like incisal surface can be seen. The spleen is shown by the white arrow.

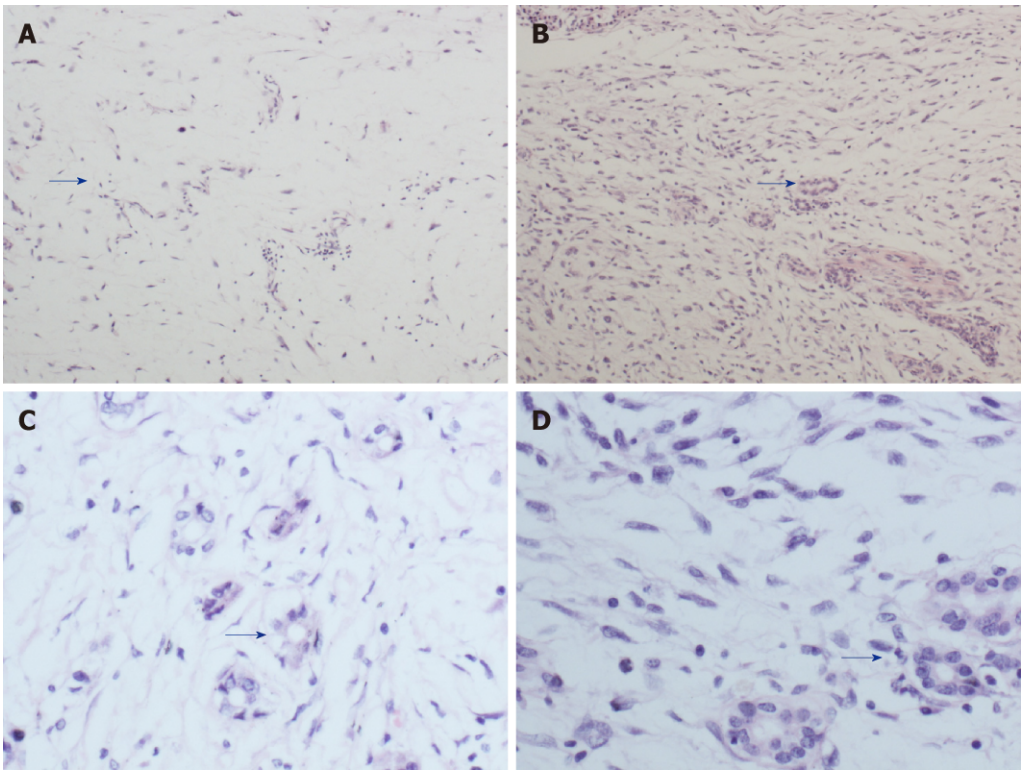

Figure 3 Histopathology. A: Coexisting areas of well-differentiated liposarcoma and the pancreatic canal (H and E × 100); B: Coexisting areas of dedifferentiated liposarcoma and the pancreatic canal (H and E × 100); C: Coexisting areas of well-differentiated liposarcoma and the pancreatic canal (H and E × 400); D: Coexisting areas of dedifferentiated liposarcoma and the pancreatic canal (H and E × 400). Blue arrows show areas containing the pancreatic canal. H and E: Hematoxylin and eosin.

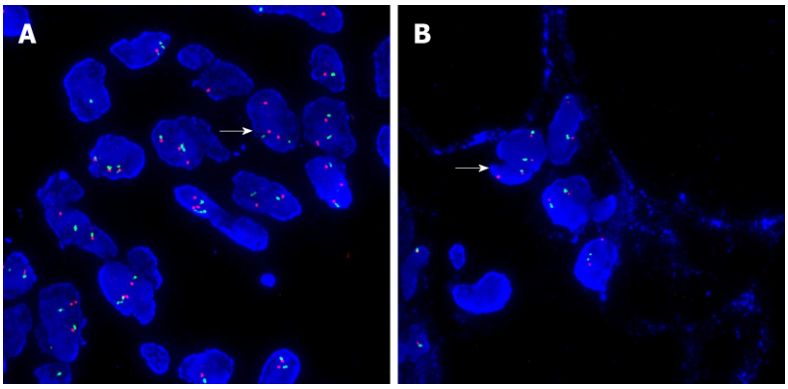

Figure 4 Fluorescence *in situ* hybridization results. A: *MDM2* gene amplification of the mass was positive (White arrow; as shown by three red dots and two green dots); B: *MDM2* gene amplification of adjacent retroperitoneal tissue was negative (White arrow; as shown by two red dots and two green dots).

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