LETTER TO THE EDITOR

Hemangioma of the pancreas in a 60-year-old woman: A report of a new case

Introduction

Few cases of pancreatic hemangioma presenting in adulthood have been documented. They are rarely suspected clinically and most are diagnosed incidentally following resection.

We report an adult pancreatic hemangioma diagnosed on pathological specimen review following distal spleno-pancreatectomy for a symptomatic mass of the body of the pancreas. This report highlights some of the difficulties encountered in clinical management.

Case report

A 62-year-old woman with a past medical history of cholecystectomy and choledocotomy for common bile duct stones. She presented to our department, 1 month later, with a 2-day history of nausea, diffuse abdominal pain, and fever. She had no other symptoms and on clinical examination, the only significant abnormal finding was epigastric tenderness. Laboratory studies were normal, particularly, amylase and lipase levels. Abdominal sonography revealed no abnormalities in biliary tract and no sign of pancreatitis on computed tomography (CT) scan; but the patient was found to have a small 2 × 1.8 cm lesion in the pancreatic body. On pre-contrast scan, the lesion appears fairly well circumscribed, homogenous with density lower than the surrounding normal pancreas. On arterial and portal phase imaging, the mass was heterogeneous, and showed a low density relative to the pancreas without marked contrast enhancement. No adenopathy was apparent, and the mass was distinct from the splenic vessels (Fig. 1). Her serum CA 19-9 was normal. Endoscopic ultrasound sonography (EUS) demonstrated the same lesion on the body of the pancreas and showed a mass with mixture of high and low echoic areas. The differential diagnosis included ductal adenocarcinoma or gastrointestinal stromal tumor. Based on CT findings, the diagnosis of typically pancreatic cystic lesion (pseudocyst, serous cystadenoma, mucinous cystadenoma..) was not evoked. Given the patient’s status, she was offered surgical resection. Intraoperatively the mass was 2 cm of diameter, included on the pancreatic parenchyma (Fig. 2). The patient underwent distal spleno-pancreatectomy. The postoperative course was uneventful, and she was discharged to home on postoperative day 10. Macroscopically, the mass measured 2.3 × 1.8 cm. It was nodular, and rela-
tively soft in consistency. On microscopic examination, the tumor showed a soft tissue mass consisted of blood vessels lined by a single layer of uniform flattened cells. Immunohistochemistry showed the spindle cells to be strongly positive for CD31 and CD34 markers, consistent with an endothelial origin. The stroma was strongly positive for actin but negative for vimentin and desmin. There was no evidence of malignancy. After 6 months of follow-up, there were no complaints or recurrence of abdominal pain.

Discussion
Pancreatic vascular neoplasms, including hemangioma, lymphangioma, hemolymphangioma, hemangioblastoma and hemangiosarcoma are non-epithelial cystic lesions of the pancreas, and according to the Kloppel’s classification, they collectively account for 0.1% of all pancreatic tumors [1]. Visceral hemangiomas have been described in various organs including the brain, parotid, thorax, liver, spleen and gastrointestinal tract. Pancreatic hemangiomas are rare. In children, pancreatic hemangiomas tend to undergo proliferation during infancy followed by a period of slow involution lasting several years and eventual regression leaving a fibrofatty residuum [2–4]. Only 10 cases of adult pancreatic hemangiomas have been reported in literature since 1939 [2]. The clinical picture and treatment usually depend on the location of the tumor and complications caused by the tumor. As in our case, most patients with pancreatic hemangioma present vague abdominal pain. In the literature, one case presented with meaena and hematemesison, and another with nausea and thrombocytopenia [2]. Preoperative diagnosis was difficult and possible malignancy could not be ruled out, and an early laparotomy was performed to make the correct diagnosis and to plan an appropriate treatment. The clinical diagnosis of this tumor is not often due to its rarity and the absence of clinical expression. Laboratory tests are frequently normal. Serum carcinoembryonic antigen (CEA) and CA 19-9 are within normal limits. Imaging techniques such as ultrasonography, CT, and magnetic resonance imaging may be used to assess, make a clinical diagnosis and for follow-up. Typically, hemangiomas are strongly contrast enhancing in the arterial phase of conventional CT [2,4]. However, cystic tumors of the pancreas often contain areas of neovascularisation and the ratio of cystic to solid tissue could influence the relative degree of tumor vascularity, so blood flow through these cavernous vascular components can be slow and this can result in diminished contrast enhancement on arterial phase [5]. As our case did not demonstrate the contrast-enhanced CT features typical of a hemangioma, Kobayachi et al. [5] and Chang et al. [6] conclude that poor arterial phase enhancement cannot rule out pancreatic hemangioma. Clinical differential diagnoses include pseudocyst, lymphangioma, sarcoma, stromal tumor, cystadenoma, cystadenocarcinoma, pseudopapillary and solid tumor, and even adenocarcinoma because of CT features. The final diagnosis is based on a combination of clinical, radiological, and histopathological findings. Pathologically, the lesion was typical of a cavernous hemangioma with blood-filled spaces. Immunohistochemistry showed like in our case, the neoplastic cells expressed the endothelial markers CD31 and CD34, and our patient is the second case of CD31 and CD34 labeling of an adult pancreatic hemangioma reported in the literature [2]. Pancreatic hemangiomas are rarely suspected clinically due to their nonspecific symptoms and most are diagnosed incidentally following resection or attempted resection for symptomatic pancreatic masses. Treatment of adult pancreatic hemangioma has been variable from laparotomy observation to subtotal pancreatectomy. Herein, we reported the first splenic distal pancreatectomy for this pathology. All cases in the literature had good prognosis, as did our case. The risk of recurrence or metastasis seems very low, but follow-up is necessary [2,4].

Conflict of interest statement
There is no conflict of interest.

References
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