

Case Report

Adult cavernous pancreatic hemangioma: a case report and review of literature

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Abstract: Pancreatic cavernous hemangioma is a benign lesion, and it is rarely seen in adults. In this case report, we analyzed the image features, including CT and MRI in a case of adult Cavernous Pancreatic Hemangioma in a 60-year-old man.

Keywords: Cavernous hemangioma, pancreatic hemangioma, computed tomography, MRI imaging

Introduction

Pancreatic vascular tumor includes hemangioma, lymphangioma, hematomylphangioma, hemangiopericytoma, hemangioblastoma and hemangiosarcoma, which only accounts for 0.1% of all pancreatic tumors [1]. Pancreatic hemangioma mostly occurs in children, which is usually associated with rapid capillary proliferation at infancy, followed by slow regression. Finally, it will vanish and leaves fibrofatty tissue [2-4]. Therefore, adult pancreatic hemangioma is rare, and only about 10 cases are reported in domestic and foreign literature. Preoperative imaging may play an important role in the correct diagnosis, help guiding clinical treatment and developing surgical plans.

Case report

A 60-year-old male patient had upper abdominal pain within unknown cause, which manifested as intermittent dull pain that was aggravated at night, with no nausea and vomiting. The symptoms were improved after taking stomach medicine, and the patient did not receive systemic treatment. Then the patient had increased pain, which could not be relieved after taking medicine. The patient had no shoulder and back issues, no yellowing of the skin nor sclera, and no skin itching. After symptom onset the patient had reduced appetite and lost 2 kg of body weight. Upon admission to the

hospital, the patient was examined and no obvious abnormalities were detected in the blood or coagulation, liver biochemistry, or tumor markers.

An upper abdomen enhanced CT (**Figure 1A**) revealed an irregular opaque soft tissue mass in the pancreas, which was lobulated, with a large layer of about 58×75 mm, the density inside the mass was uneven, and patchy low density opacity could be observed around the edge. Enhanced scanning (**Figure 1B-D**) revealed no obvious enhancement, and the lesion was ill-defined within the descending and transverse duodenum. Further abdominal MRI enhancement revealed that the tumor showed hypointensity on T1WI, and slight hyperintensity opacity on T2WI, as well as patchy hyperintensity on the edge with T1WI and T2WI (**Figure 1E, 1F**), the pancreatic duct was slightly dilated, enhanced scanning revealed no obvious enhancement (**Figure 1G**).

Intraoperative observation suggested that, the tumor was located at the uncinate process of the pancreatic head, which was ill-defined, with a size of about 60×50 mm; and about 20 cm mesentery in the transverse and ascending duodenum, as well as the initial segment of jejunum which was thickened, with the thickness of about 5 cm. The possibility of a malignant tumor could not be excluded, and therefore pancreaticoduodenectomy was performed.

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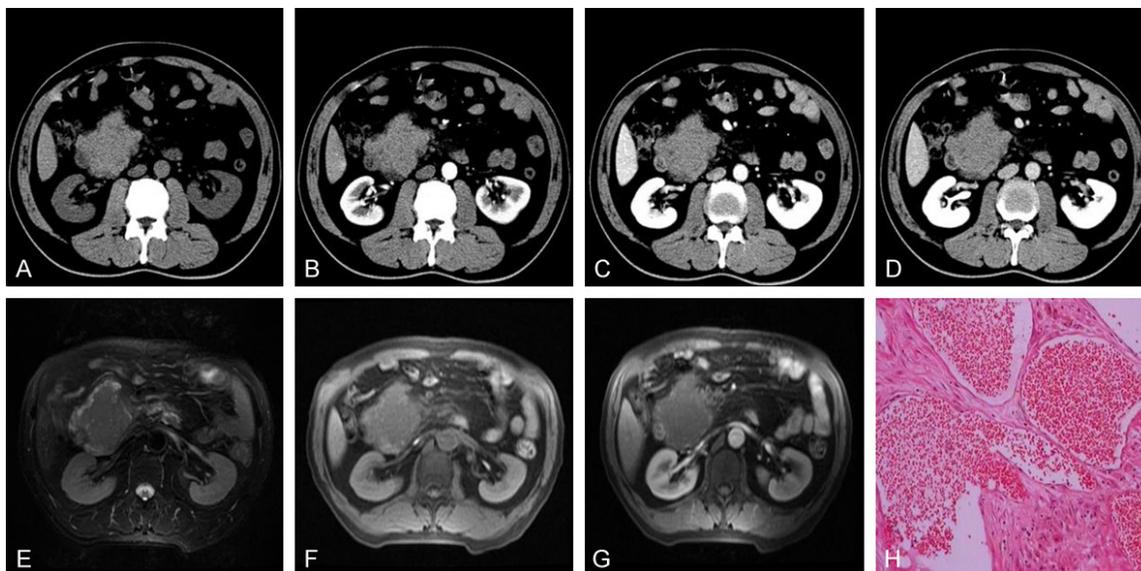


Figure 1. A. CT revealed a lobulated soft tissue mass between the pancreatic head and the duodenum, which was uneven in density, with the CT value of about 50 HU, and patchy low density opacity could be seen on the lesion edge, which was ill-defined. B-D. Showed the arterial phase (51 HU), venous phase (54 HU) and delayed phase (55 HU) of enhanced scanning, and the tumor showed no obvious enhancement. E, F. The tumor displayed slight hyperintensity on T2WI and hypointensity on T1WI, patchy hyperintensity on T2WI and isointensity on T1WI could be seen on the lesion edge. G. Enhanced scanning showed no obvious enhancement of the lesion. H. Microscopic observation (H&E staining $\times 100$).

Postoperative pathology (**Figure 1H**) indicated that the pancreas conformed to the cavernous hemangioma, among which, the majority showed angiolipoma changes, while the (mesentery) local vessels and adipose tissues showed tumor-like hyperplasia changes.

Discussion

Adult pancreatic hemangioma is mostly seen in adult females, which grows slowly, with no specific clinical manifestation. Typically, the patient may show no symptoms when the tumor is small in volume, but when the tumor grows to a certain degree, it may manifest as abdominal pain discomfort or large abdominal mass, or jaundice when it has compressed the common bile duct or duodenum. Large hemangiomas with rich blood vessels may rupture or bleed due to trauma or strenuous activity; alternatively, the increased blood supply in the tumor body as a result of intra-abdominal pressure elevation due to pregnancy or hemodilution in the late trimester of pregnancy may also induce bleeding [5].

In the clinic, hemangioma is classified as cavernous hemangioma, capillary hemangioma, cirroid hemangioma and mixed hemangioma, among which, cavernous hemangioma is the

most commonly seen. Under microscopic observation, the tumor is constituted by small blood vessels or blood sinus filled with blood and organized thrombus, with a monolayer lining of vascular endothelial cells, and there is a cavernous structure formed by the fibrous septum. Positive vascular endothelial markers CD31, CD34 and FVIII-RAg, while negative lymphatic endothelial marker D2-40 in immunohistochemistry confirms the diagnosis.

Cavernous hemangioma is frequently seen in the liver, which is associated with the typical manifestations of clear nodular enhancement of lesion edge at arterial phase, and gradual inward filling of the enhancement at venous phase and delayed phase. The mode of enhancement of pancreatic cavernous hemangioma is not the same as that of hepatic hemangioma, and only 1 case report suggests that the lesion shows obvious enhancement [6]. The pancreatic cavernous hemangiomas reported in the remaining domestic and foreign literature are mostly cystic and solid, while a minority are solid, with no obvious enhancement or mild enhancement. Some scholars believe that, the absence of obvious tumor enhancement may be related to the fact that the microstructure of pancreatic hemangioma is different from that of hepatic hemangioma, part of the blood ves-

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sels and blood sinuses inside the tumor are not connected to the blood circulation of patients, since histological examination shows that these blood sinuses contain plasma rather than blood [7]. The research results by Lee et al. further supported such hypothesis [8], in which no blood flow signal was detected in color doppler ultrasonography. Moreover, some scholars propose that the absence of obvious tumor enhancement is the result of the fact that there is newly formed blood vessel with artery-vein shunts within the tumor, and the blood flow slows down when passing these regions, which thereby weakens the lesion enhancement at the arterial phase [9]. Additionally, the cystic and solid component ratio in the tumor will also affect the relative degree of tumor vascular distribution, thus impacting the lesion enhancement degree at the arterial phase [10]. Therefore, the possibility of hemangioma cannot be excluded in the absence of clear lesion enhancement at arterial phase of enhanced scanning. According to literature reports, the pancreatic cavernous hemangioma is mostly well-defined. However, in this case, the tumor was ill-defined with the surrounding duodenum, CT findings suggested patchy low density opacity on the lesion edge, while MRI findings revealed patchy hyperintensity on T2WI and isointensity on T1WI. In addition, the pathology revealed tumor-like hyperplasia changes in the local blood vessels and adipose tissue of the mesentery, which were consistent with the imaging findings.

Pancreatic cavernous hemangioma is a benign lesion, and patients can have regular clinical visits if they are asymptomatic; however, the lesion is associated with risk of bleeding, and it is difficult to diagnose before surgery, so the possibility of a malignant lesion cannot be excluded. Therefore, cases reports in the literature are mostly verified through surgical pathology. In this case, the patient had the preoperative manifestation of upper abdominal pain discomfort, and a retroperitoneal malignant lesion could not be excluded through clinical and imaging examinations, so pancreaticoduodenectomy was carried out. The pathology conformed to pancreatic cavernous hemangioma, and no tumor recurrence or metastasis was observed after 5 years of follow-up after surgery.

In summary, pancreatic cavernous hemangioma is mostly cystic and solid, while a minority is

solid, enhanced scanning usually shows no obvious enhancement or only mild enhancement. The tumor grows slowly, and negative tumor markers contribute to the differential diagnosis, but a definite diagnosis should be made based on pathological examination.

Disclosure of conflict of interest

None.

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