Case Report Pancreatic duct stones misdiagnosed as intraductal papillary mucinous neoplasms of the pancreas: a case report

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Abstract: Intraductal papillary mucinous neoplasms (IPMN) of the pancreas are a kind of mucin-producing tumor and have gained great epidemiological and clinical recognition because of the widespread use of cross-sectional abdominal imaging. The wide application of imaging also has led to a surge in the number of pancreatic cysts incidentally diagnosed. In this article, we report a case of pancreatic duct stones which was misdiagnosed as IPMN. A 62-year-old male with abdominal pain was diagnosed with IPMN before the operation, but the final postoperative pathology examination showed that he really had chronic pancreatitis with pancreatic duct stones. In this case, the magnetic resonance cholangiopancreatography (MRCP) suggested there was a cystic mass in the head of the pancreas, accompanied by a dilatation of the intrahepatic and extrahepatic bile ducts and the main pancreatic ducts (> 10 mm). A nodular, low-signal mass was seen with T2-weighted imaging (T2W1) in the head of the pancreas, and it was similar to the magnetic resonance imaging (MRI) manifestation of the pancreatic duct stones. The postoperative course was uneventful. The patient was discharged on postoperative day seven and followed up regularly after discharge. Up to now, there has been no sign of recurrence, and his blood glucose is controlled well. This case gives us some warnings, and the realization that relying on only one imaging examination can easily lead to a misdiagnosis. The flexible use of multiple imaging examinations can contribute to the accurate diagnosis of the disease.

Keywords: Abdominal pain, pancreatic intraductal papillary mucinous neoplasms, chronic pancreatitis, pancreatic duct stones, diagnosis

Introduction

Mucin-producing tumors of the pancreas are most commonly divided into mucinous cystic neoplasms (MCN) and intraductal papillary mucinous neoplasms (IPMN). They are a group of premalignant or malignant neoplasms forming multilocular cysts and are lined with tall, columnar mucinous epithelia [1]. IPMN of the pancreas was first reported by Ohashi et al. in 1982 [2]. It mainly occurs in older males, with a median age of about 60-70 years [3, 4]. There are many ultrasonographic and abdominal cross-section imaging examination approaches, such as computed tomography (CT) scans, MRI, and endoscopic ultrasonography (EUS), for diagnosing IPMN. These approaches have resulted in a greatly increased clinical recognition of IPMN because of the widespread application of abdominal cross-section imaging, which leads to a sharp increase in the number of pancreatic cysts diagnosed [2]. The diagnostic rate of IPMN is increasing along with the development of imaging examinations and laboratory tests, but misdiagnoses are still unavoidable. Clinically, the diagnosis of IPMN is often delayed due to its similarity to pancreatitis or cystic tumors of the pancreas [5]. However, in this report, we present the case of a 62-yearold male who was diagnosed with IPMN before his operation, while the pathology suggested he had chronic pancreatitis with pancreatic duct stones. This study was approved by the Ethics Committee of the Affiliated Hospital, Southwest Medical University. The patient gave a written informed consent for his participation in the study.

Case report

The patient was a 62-year-old male who had a tolerable but persistent burning pain in the upper abdomen for more than one year. The accompanying symptoms were acid reflux and



Figure 1. The MRCP imaging of the patient: A: There was a cystic mass in the head of the pancreas with a long T1, a long T2 signal, and a low signal in DWI. The maximum cross-section was about 1.7*1.1 cm. B: The mass (the red arrow) was linearly separated from the main pancreatic duct (the yellow arrow). C: The adjacent common bile ducts were compressed and narrowed (the red arrow). The main pancreatic ducts were dilated (the yellow arrow).

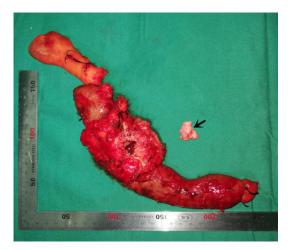


Figure 2. The patient's resection specimen: There was a 1.5*1.5 cm white pancreatic stone (the black arrow) in the head of the pancreas when we cut it open.

belching. Taking some drugs brought considerable relief from the pain, but the medication did not make the pain disappear. He lost about 5 kg of weight during his illness. The patient had a history of tuberculosis but was only treated for two months. His physical examination was normal. As for the laboratory tests, his liver function test showed that his ALT (alanine aminotransferase) level was 152.8 U/L (9-50 U/L), his AST (aspartate aminotransferase) level was 84.5 U/L (15-40 U/L), his TBIL (total bilirubin) level was 26.4 umol/L (0-23 umol/L), his DBIL (direct bilirubin) level was 24.0 umol/L (0-7 umol/L), and his tumor markers showed that his CEA (carcinoembryonic antigen) level was 11.72 ng/ml (0.00-6.00 ng/ml), and his FER level was 433.58 ng/ml (25.00-280.00 ng/ml). His blood clotting test was normal. The regular monitoring of his blood glucose after admission found that his blood sugar level was increased but was well controlled by insulin.

The gastroscopy revealed chronic non-atrophic gastritis with erosions. The abdominal B-ultrasound suggested there were stones in the inferior segment of the common bile duct with intrahepatic and extrahepatic bile duct dilatation and main pancreatic duct dilatation. A further enhanced MRI and MRCP of the upper and middle abdomen suggested there was a cystic mass in the head of the pancreas with a long T1 signal, a long T2 signal, and a low signal in DWI. Its maximum cross-section was about 1.7*1.1 cm (Figure 1A). The cyst wall was markedly enhanced and linearly separated from the main pancreatic duct (Figure 1B). The cystic mass seemed to be partially connected with the main and accessory pancreatic ducts. The adjacent common bile ducts were compressed and narrowed (Figure 1C), and the main pancreatic duct and the intrahepatic and extrahepatic bile ducts were dilated. The wall of the distal common bile duct was thickened and strengthened.

According to the imaging features and the clinical manifestations, the patient was diagnosed with probable IPMN of the pancreas. IPMN is a premalignant tumor, and the patient had some high-risk features such as abdominal pain and a main duct dilation \geq 10 mm, so the patient accepted the operation (Whipple). During the operation, it was found that the diameter of his common bile duct was about 10 mm, and the head and uncinate process of the pancreas were as hard as stone. After the operation, there was a 1.8×1.5 cm white pancreatic stone found in the head of the pancreas when we dissected the tissue (Figure 2). A postoperative pathological examination revealed there was a hard area of 3.5×3×2 cm in the pancreas specimen, and it was gravish white with a partial cystic change. The pancreatic duct showed cystic dilatation with stones. Under the microscope,

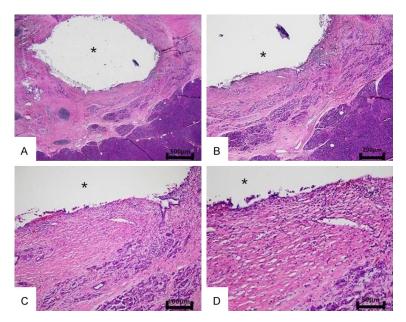


Figure 3. Pathological section of the patient: The pancreatic duct was cystically dilated, the common pancreatic duct was dilated with the formation of the stones, and there were inflammatory changes in the surrounding pancreatic tissue. (A. Bar = $500 \mu m$; B. Bar = $200 \mu m$; C. Bar = $100 \mu m$; D. Bar = $50 \mu m$; * Pancreatic duct).

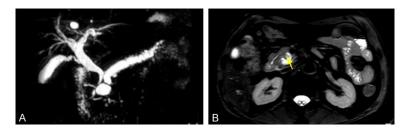


Figure 4. A (The MR hydrography 3D imaging): A cystic mass is located in the head of the pancreas, accompanied by a dilatation of the intrahepatic and extrahepatic bile ducts and the main pancreatic ducts, especially the main pancreatic duct. B: A nodular low-signal mass (the yellow arrow) and high-signal dilated pancreatic ducts with T2WI were seen in the head of the pancreas.

we could see the recognizable abnormalities, including a thickening of the pancreatic duct wall, fibrosis, inflammatory cell accumulation, inflammatory changes in the surrounding pancreatic tissue (**Figure 3**). According to the pathological findings, we determined that the patient had chronic pancreatitis with pancreatic duct stones, not IPMN. The preoperative MRCP report and the clinical diagnosis were not consistent with the postoperative pathological diagnosis.

Discussion

IPMNs secrete a large amount of mucoid substances which accumulate in the main pancre-

atic duct or branch pancreatic duct, resulting in the corresponding pancreatic duct dilatation [6]. According to the extent to which they involve the pancreatic ductal system, IPMNs of the pancreas are primarily divided into the main duct (MD-IPMN) type, the branch duct (BD-IPMN) type, and the mixed duct type [7, 8]. The MD-IPMN is characterized by a diffuse or partial dilatation and the main pancreatic duct (> 5 mm) filled with excessive mucin. This type occurs mainly at the head of the pancreas and occasionally at the tail of the pancreas. BD-IPMN affects one or more branches of the pancreatic duct, showing cystic dilatation (> 5 mm), and the mixed-type IPMN is a combination of the MD-IPMN and BD-IPMN [2, 7]. Common symptoms of IPMNs include epigastric discomfort or pain (70-80%), nausea and vomiting (11-21%), backache (10%), weight loss (20-40%), diabetes, and jaundice [7].

In this case, IPMN of the pancreas cannot be easily differentiated from chronic pancreatitis (CP) through the clinical symptoms such as abdominal pain, high blood sugar, and weight loss, and, furthermore, the long history of smoking

and drinking which this patient has is a common high-risk factor for IPMN of the pancreas and chronic pancreatitis [9]. The symptoms and the laboratory tests suggest that the pathogenesis may be located in the pancreas, so we need to rely on the imaging examination for a further diagnosis.

As for the imaging examination, the B-ultrasound suggested there were stones in the common bile duct and no abnormality in the pancreas, but the MRI found a mass in the pancreas. In the MR hydrography 3D imaging (**Figure 4A**) of this case, we observed that a cystic mass was located in the head of the pancreas, accompanied by a dilatation of the intrahepatic and extrahepatic bile ducts and the main pancreatic ducts (> 10 mm), especially the main pancreatic duct, which is similar to the diffuse dilatation of the main pancreatic duct type IPMN. After the operation, we reviewed carefully the MRI image and found that the nodular low-signal mass and high-signal dilated pancreatic ducts with T2WI were seen in the head of the pancreas (Figure 4B). This is similar to the MR imaging of type 1 pancreatic duct stones [10]. Computed tomography (CT) is considered to be the best initial imaging test for CP and CP with pancreatic duct stones [10, 11]. The absence of the CT examination may be the main cause of the misdiagnosis in this case. Thus, through this case, we believe that two or more imaging examinations are needed for the diagnosis and differential diagnosis of a disease.

Various cross-sectional and ultrasonographic imaging methods are used in the assessment and observation of patients with IPMN, and each has its own advantages and limitations. CT has been reported as an effective method for diagnosing BD-IPMN. It can detect calcification well and evaluate the nodule, septum and wall thickening [12, 13]. Harima et al. [14] reported that using CT to detect mural nodules in BD-IPMN produced a sensitivity, specificity and accuracy of 68%, 100%, and 77%, respectively. Meanwhile, contrast-enhanced CT has a good ability to predict malignant IPMN. In the study of Lee et al. [15] which included 83 cases of IPMN of the pancreas, and according to the revised 2017 international consensus guidelines, the sensitivity and specificity of contrastenhanced CT in predicting malignant IPMN were 86% and 74%, respectively. In a word, CT is very helpful for the diagnosis and the differentiation of benign or malignant IPMN of the pancreas, especially in combination with enhanced scanning. As for MRI, because of the high resolution of MRI with soft tissue, it can display the morphological details of IPMN and the internal structure of cystic masses more clearly than CT, and DWI has been proved to be an effective method for diagnosing malignant IPMN [16-18]. Meanwhile, MRCP is a reliable measure for determining cyst location, size, the dilation of the pancreatic duct, the pancreatic duct system, and the communication with the main pancreatic duct [2, 8, 19].

The cystic cavity communicating with the main pancreatic duct is a reliable sign for the diagno-

sis of IPMN of the pancreas. The display rate of IPMN communicating with the pancreatic duct using MRCP is significantly higher than with CT [8, 19], and the value of enhanced MRI, enhanced CT and EUS in predicting the malignant potential of IPMN is nearly equal [15, 20]. In fact, the resolution ratio and sensitivity of MRI are not sufficient for diagnosing patients with early-stage IPMN [8, 19], but in a recent study by Sun et al. [19], they suggested that the combination of MRI and contrast liposome composed of target molecules of IPMN and Gd-EOB-DTPA can be used to diagnose patients suspected of having IPMN. In any case, MRI is still the preferred imaging modality for the follow-up of pancreatic IPMN. First, MRI does not make use of radiation, so it is a safer method for patients who need long-term monitoring. Second, MRI imaging for pancreatic cysts does not require contrast, so even nocontrast MRI can detect the undesirable changes [21].

EUS is another common method of diagnosing IPMN. The main advantage of EUS is to evaluate the mural nodules of IPMN. It's reported that as much as 28% of CT and MRI misses the diagnosis of mural nodules in patients with BD-IPMN compared with EUS [14]. But compared with the CT and MRCP, EUS is a more invasive diagnostic procedure, and most patients would have undergone prior abdominal imaging before EUS generally, so the EUS is often performed as part of a multi-modality diagnostic evaluation [22].

Conclusion

MRI is equivalent to CT in the diagnosis of IPMN and the differentiation of benign or malignant of IPMN. In view of the advantage of MRI without radiation, MRI can be selected preferentially if the patient can afford it. It is also the first choice for conservative observation or postoperative follow-up. Although MRI is the optimal choice for a noninvasive IPMN examination, it is not the only examination. What we learned from this case is that we should use two or more diagnostic imaging modalities to diagnose the disease according to the actual situation.

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Disclosure of conflict of interest

None.

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