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Case Report

Misdiagnosis of Anterior Superior Pancreaticoduodenal Artery Aneurysm Rupture Likely Due to Segmental Arterial Mediolysis : A Case Report

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Abstract : An aneurysm of the abdominal internal organs is relatively rare. Recently, segmental arterial mediolysis (SAM) and median arcuate ligament syndrome (MALS) were identified as specific causes for aneurysms of the pancreaticoduodenal artery arcade. Herein, we report a ruptured anterior superior pancreaticoduodenal artery (ASPDA) aneurysm due to SAM that was misdiagnosed as acute pancreatitis. The patient was a 59-year-old male with acute, severe, and sharp pain in the upper abdomen. He was clinically diagnosed with acute pancreatitis based on abdominal computed tomography (CT). However, a follow-up CT scan revealed an aneurysm of the ASPDA. We therefore diagnosed this case as retroperitoneal hemorrhage due to aneurysm rupture, and we performed an angiogram and transcatheter arterial embolization to prevent aneurysm re-rupture. Based on a subsequent review of all the findings for this patient, we retrospectively determined the cause of the ASPDA aneurysm to be SAM. Such case reports are rare, and further accumulation of similar cases is necessary in the near future to establish proper diagnostic criteria and appropriate treatment protocols.

Key words : SAM, aneurysm, ASPDA, misdiagnosis, pancreatitis

Introduction

Aneurysms of the abdominal internal organs are relatively rare, although such occurrences could be fatal. In symptomatic cases, abdominal aneurysm requires prompt diagnosis and treatment[1–3]. Segmental arterial mediolysis (SAM) and median arcuate ligament syndrome (MALS) were recently demonstrated as specific causes of aneurysm in the pancreaticoduodenal artery arcade[4–7]. Here, we present a case of retroperitoneal hematoma due to the rupture of an anterior superior pancreaticoduodenal artery (ASPDA) aneurysm that was first misdiagnosed as acute pancreatitis. The patient was eventually treated successfully by transcatheter arterial embolization.

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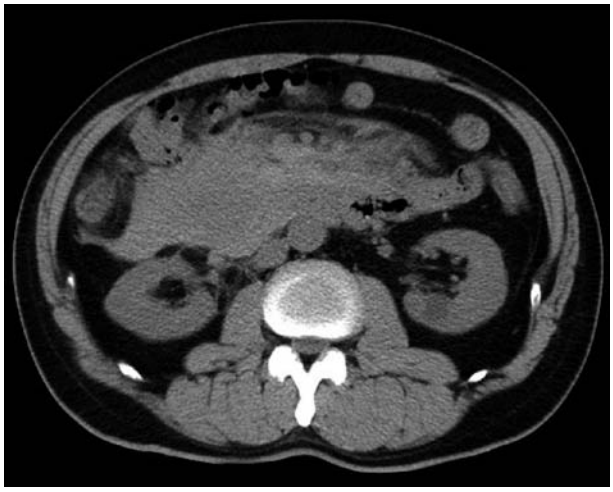

Fig. 1. Plain CT showing fluid retention in the anterior pararenal extraperitoneal space and around the pancreas head.

Case Report

A 59-year-old man was referred to our hospital with acute, severe, and sharp pain in the upper abdomen. He was a habitual daily drinker, but had no history of pancreatitis. Laboratory tests revealed normal levels of amylase, calcium, and triglyceride, and an increased white blood cell count. A plain computed tomography (CT) scan of the abdomen showed fluid retention mainly in the anterior pararenal extraperitoneal space, involving the head and body of the pancreas (Fig. 1). The patient was clinically diagnosed with acute pancreatitis, hospitalized, and treated with intravenous feeding management and intravenous antibiotics. The next day, an enhanced CT scan showed CT grade 1 fluid retention around the pancreas head that was not visible without enhancement. Therefore, we diagnosed acute pancreatitis with no severity, based on the Japanese guidelines of acute pancreatitis[8, 9]. A follow-up CT scan on the 8th day after admission showed increased fluid retention, local expansion and stenosis of the artery, and an aneurysm (approximately 10 mm in diameter) of the ASPDA that was not detected on the previous CT (Fig. 2). Other blood vessels were normal and there was no intestinal tract ischemia. At this time, laboratory tests showed no abnormal data. Therefore, we diagnosed this case as retroperitoneal hemorrhage due to an aneurysm rupture rather than acute pancreatitis. Although the patient's abdominal pain was subsiding, we performed angiogram and transcatheter arterial embolization to prevent aneurysm re-rupture (Fig. 3). There was no stenosis in the root of the celiac artery or superior mesenteric artery, and good blood flow was maintained. Furthermore, the ASPDA branched from the gastroduodenal artery, and the aneurysm was located and found to have a central diameter of 10 mm. Dilatation and constriction were treated with contrast media to continuously link the corresponding CT findings. Subsequent superior mesenteric arteriography showed enhancement of the common hepatic artery, left gastric artery, and splenic artery in the area of the celiac artery through the pancreaticoduodenal artery

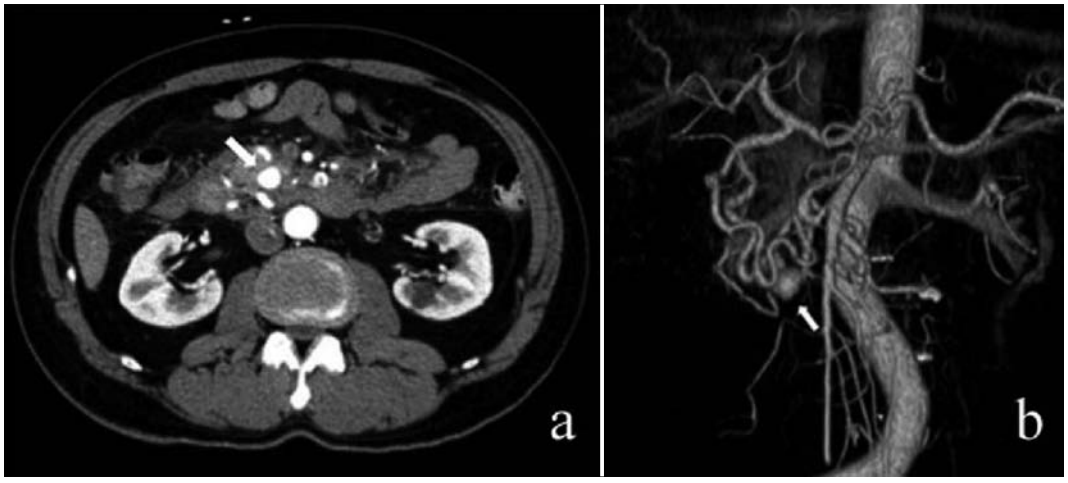

Fig. 2.
a : Enhanced CT showing an ASPDA aneurysm of approximately 10 mm in diameter (white arrow).
b : 3D-CT revealing an ASPDA aneurysm (white arrow).

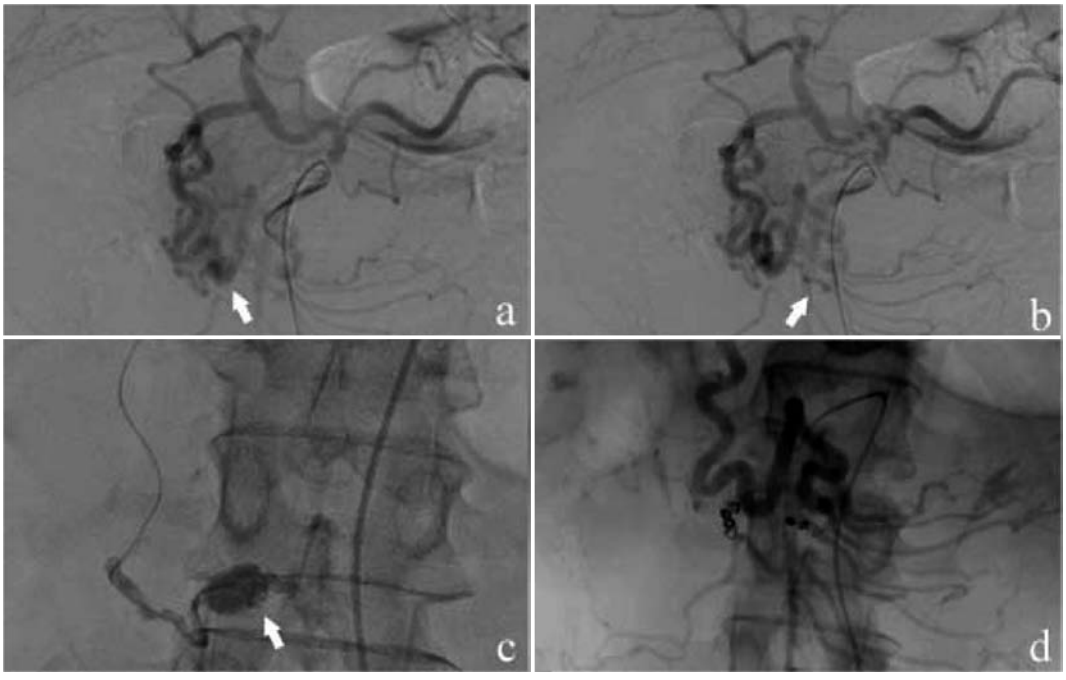

Fig. 3. Celiac angiogram demonstrating an ASPDA aneurysm
(a) (white arrow) and continuously repeating dilatation and constriction (b) (white arrow). Before (c) (white arrow indicates the aneurysm) and after (d) TAE. The aneurysm was not completely visible.

arcade, suggesting compression in the root of the celiac artery. Consequently, both ends of the aneurysm were embolized using coils. For some days thereafter, biliary vomiting continued due to compression of the duodenum by hematoma ; therefore, we inserted a nasogastric tube. On the 24th day, a repeat CT revealed reduced hematoma, and on the 32nd day, duodenography

through the nasogastric tube demonstrated outflow of contrast medium to the anal side. The patient was discharged on the 38th day and remains in good health.

Discussion

An abdominal aneurysm is relatively rare[1], with reported incidences of 60% in the splenic artery, 20% in the hepatic artery, 5～8% in the superior mesenteric artery, 4% in the celiac artery, and 2% in the pancreaticoduodenal artery[10, 11]. Moreover, stenosis of the celiac artery is observed in 68～74% of cases of pancreaticoduodenal artery aneurysm, while such aneurysm is observed in 80% cases of celiac arterial stenosis[12]. Abdominal aneurysms are generally attributed to an infection, such as infectious mononucleosis, pancreatitis, arteriosclerosis, tumor invasion, or celiac stenosis. In recent years, SAM and MALS were suggested as additional likely causes[4–7]. There are no specific symptoms from a ruptured aneurysm in the pancreaticoduodenal artery; patients show general clinical symptoms including abdominal pain, back pain, gastrointestinal bleeding, jaundice, and general malaise[13]. Various imaging tests are useful for diagnosing an aneurysm rupture, and many hematomas are revealed by abdominal enhanced CT examination with confirmation by angiography. In the present case, it took several days to diagnose the retroperitoneal hematoma, following an initial diagnosis of acute pancreatitis. Hindsight indicated that he was misdiagnosed because the initial clinical symptoms and CT imaging were very similar to those observed in patients with acute pancreatitis. However, the CT value of the abdominal mass was approximately 50 HU, and it was clearly different from the effusion characteristic of pancreatitis. More careful attention to this CT value at the time might have facilitated the detection of hematoma. Furthermore, blood biochemical tests demonstrated no increase in serum amylase, and we could not check the serum lipase recommended in the guidelines[9]. Retrospectively, there was little evidence to confirm acute pancreatitis in this case. We finally diagnosed this case as SAM, which is very difficult to diagnose prior to bleeding and without the appropriate histopathological examinations[14–18]. In this case, the aneurysm could be resolved via embolization without surgery.

Uchiyama *et al*[19] established the clinical diagnostic criteria of SAM as follows: a) middle-aged and elderly individuals, b) no underlying disease such as inflammation or arteriosclerosis, c) sudden intra-abdominal hemorrhage, and d) irregular extension and stenosis (beading and narrowing) on angiography. Michael *et al*[20] also reported that it is possible to diagnose SAM using angiography or CT if vasculitis is ruled out. The present case was diagnosed as SAM according to these criteria. Furthermore, a similar case report of SAM was originally misdiagnosed as acute pancreatitis based on clinical symptoms and imaging findings of retroperitoneal bleeding[21].

MALS is a chronic stenosis of the celiac artery due to the compression of the median arcuate ligament. In this condition, blood to the liver and spleen is supplied through the pancreaticoduodenal artery arcade from the superior mesenteric artery. As a result, pancreaticoduodenal artery aneurysm is formed due to hemodynamic stress on the artery[7]. Although the incidence of aneurysm from this disease is unknown, Quandalle *et al*[22] reported

stenosis or occlusion of the celiac artery in 23 of 34 cases of pancreaticoduodenal artery aneurysm. In the present case, stenosis of the celiac artery was unclear by CT and arteriography, thus MALS could be discounted. A key point in the diagnosis of pancreaticoduodenal artery aneurysm is therefore the findings on CT or angiography of non-specific symptoms. Moreover, it is important to differentiate SAM and MALS from other causes.

We recommend arterial catheter treatment as the first choice in cases such as the one reported herein because it is less invasive than surgery. However, surgery may be required in specific circumstances.

Conclusion

We reported a case of rupture to an ASPDA aneurysm due to SAM, with a misdiagnosed retroperitoneal hematoma first diagnosed as acute pancreatitis. Although SAM and MALS are possible causes of abdominal aneurysm, the reported cases are rare. An accumulation of cases, the establishment of diagnostic criteria, and an appropriate treatment strategy will be necessary in the near future.

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Conflict of interest

We declare that there are no conflicts of interests.

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