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CASE REPORT

Hyper-dense fluid on plain computed tomography may reveal a ruptured aneurysm in patients with median arcuate ligament syndrome

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Abstract
Rupture of abdominal aneurysms associated with median arcuate ligament syndrome (MALS) is a serious condition and requires accurate diagnosis in a clinical setting. We examined three patients with this condition: two women aged 45 and 71 years, and a 61-year-old man. They complained of abdominal pain and had fluid collection around the duodenum. Plain computed tomography (CT) of the fluid collection revealed hyper density, which suggests the presence of blood. Moreover, contrast-enhanced CT revealed aneurysms in the pancreatic head area. Angiography revealed aneurysms of the branches of the gastroepiploic artery, which were treated by endovascular embolization in all patients. Thus, patients with abdominal pain and high-density fluid collection around the duodenum on plain CT may suffer from hemorrhage following rupture of MALS-associated aneurysms.

Keywords Median arcuate ligament syndrome · Aneurysm · Plain computed tomography

Introduction

Median arcuate ligament syndrome (MALS) is a disorder wherein the root of the celiac artery is compressed by the median arcuate ligament [1]. The blood flow through the celiac artery is decreased and that through the hepatic and splenic artery is provided by the superior mesenteric artery via the pancreatic head arterial arcade. The increased blood flow through the arcade elevates the pressure in the arteries and causes aneurysm [2].

Rupture of aneurysms cause acute abdominal pain with fluid collection around the pancreas and the duodenum. This condition mimics that of acute pancreatitis, but serious cases require emergent treatment by endoscopic coiling [3] to prevent hemorrhage-induced hypotension. For an accurate diagnosis, imaging modalities are crucial.

Here, we report the cases of three patients with ruptured aneurysms in the pancreatic head arcade with MALS. Plain computed tomography (CT) accurately diagnosed hemorrhage of the aneurysm in these patients.

Patient 1

A 45-year-old woman was admitted to the emergency department of our hospital because of acute abdominal pain with hypotension (83/50 mm Hg) and anemia (HB = 9.3 g/dl). Plain CT revealed a hyper-dense fluid collection, suggestive of blood presence, around the second portion of the duodenum, which extended into the peritoneal cavity (Fig. 1a). On contrast-enhanced (CE)-CT, an aneurysm was revealed in the fluid collection (Fig. 1b). CT angiography (AG) revealed a narrowing at the root of the celiac artery and an aneurysm in the pancreatic head arcade (Fig. 1c). AG of the superior mesenteric artery revealed an aneurysm at the confluence of the branch of the dorsal pancreatic artery and anterior pancreaticoduodenal artery. The common hepatic artery and the splenic artery were opaque, reflecting a decreased blood flow through the celiac artery (Fig. 1d). Although AG did not reveal extravasation from the aneurysm, rupture of the aneurysm with MALS was considered; hence, the aneurysm was treated by transcatheter arterial embolization (TAE) with coils. Following treatment, hemostasis was achieved,

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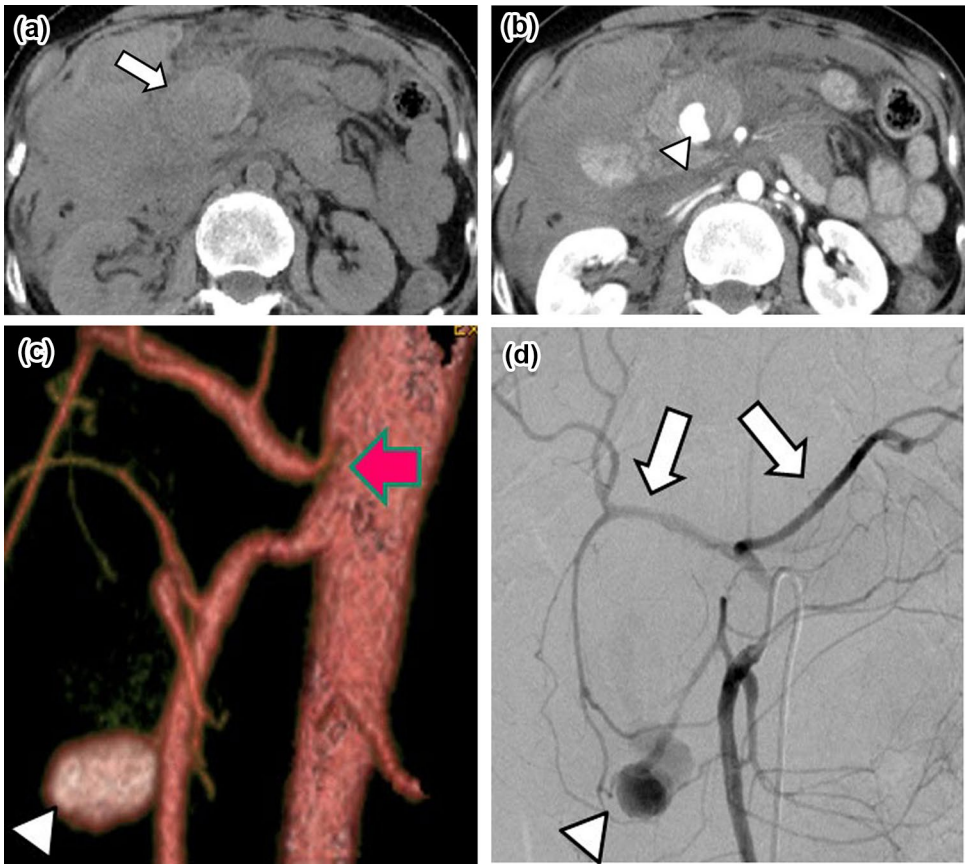

Fig. 1 Case 1. **a** Abdominal plain computed tomography (CT). A hypo-dense lesion is revealed around the second portion of the duodenum extending into the peritoneal cavity. The hyper-dense region is included, suggesting that the lesion is a hematoma (arrow). **b** Abdominal contrast-enhanced CT. The contrast medium is accumulated within the lesion (arrow head). **c** CT angiography (AG). An aneurysm is revealed in the pancreatic head arcade area (arrow head) with a narrowed root of the celiac artery (arrow). **d** AG through the superior mesenteric artery. The common hepatic artery and the splenic artery are opacified (arrows) with the aneurysm at the confluence of a branch of the dorsal pancreatic artery and the anterior pancreaticoduodenal artery (arrow head)

and no recurrence of aneurysm was observed for 6 years after treatment.

Patient 2

A 61-year-old man was admitted with epigastralgia that had lasted for 10 days. Blood examination did not reveal anemia (HB = 14.5 g/dl). Plain CT revealed a high-density fluid collection around the second duodenal portion (Fig. 2a), CE-CT revealed an aneurysm in the fluid collection without extravasation (Fig. 2b), and CT-AG revealed that the aneurysm was located in the anterior superior pancreaticoduodenal artery (Fig. 2c) and that a narrowing was located at the root of the celiac artery (Fig. 2d). Magnetic resonance imaging T1- and T2-weighted images revealed a high intensity of the fluid collection. AG of the superior mesenteric artery revealed an aneurysm of the anterior superior pancreaticoduodenal artery without extravasation and an opacification of the common hepatic artery and the splenic artery (Fig. 3a). The aneurysm was treated by TAE with coils. Following treatment, hemostasis was achieved. Esophagogastroduodenoscopy (EGD) revealed a narrowing of the second portion of the duodenum 7 days after TAE (Fig. 3b). The narrowing of the second portion of the duodenum, presumably due to hematoma, was relieved after treatment on EGD conducted 20 days after TAE (Fig. 3c). No recurrence of aneurysm was observed for 2 years following treatment.

Patient 3

A 71-year-old woman was admitted with upper abdominal pain that had lasted for 8 days. Plain CT revealed fluid collection with high density around the second duodenal portion (Fig. 4a). Blood examination indicated slight anemia (HB = 12.1 g/dl). CE-CT revealed an aneurysm adjacent to the pancreatic head (Fig. 4b), and CT angiography revealed a narrowing at the root of the celiac artery (Fig. 4c). AG of the celiac artery did not completely opacify the common hepatic artery and the splenic artery; however, clear opacification of these arteries was revealed on AG of the superior mesenteric artery. Moreover, an aneurysm of the inferior pancreaticoduodenal artery without extravasation was recognized (Fig. 4d); therefore, endovascular embolization was performed with coils. Following treatment, hemostasis was achieved, and no recurring aneurysm was observed for 8 months following treatment.

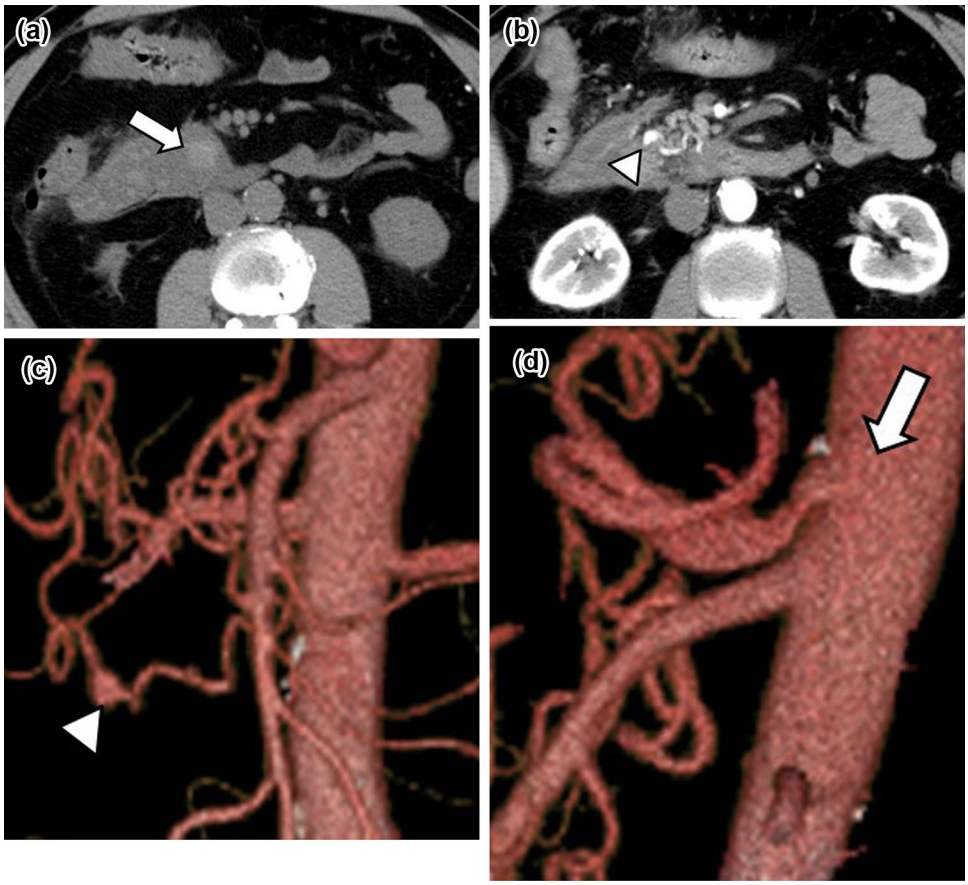

Fig. 2 Case 2. **a** Abdominal plain CT. A hypo-dense lesion is revealed around the second portion of the duodenum. A hyper-dense area can be observed, suggesting that the lesion is a hematoma (arrow). **b** Abdominal CE-CT. The contrast medium is accumulated within the lesion (arrow head). **c** CT-AG. An aneurysm of the anterior superior pancreaticoduodenal artery is visible (arrow head). **d** CT-AG. A narrowed root of the celiac artery is also visible (arrow)

Discussion

An abdominal visceral aneurysm is a rare condition with an incidence rate of 0.1–0.2% of all reported aneurysms [4]. Among patients with abdominal visceral arteries, the splenic artery is most frequently afflicted (50%), followed by the hepatic artery (20%), superior mesenteric artery (8%), celiac artery (4%), pancreaticoduodenal artery (2%), renal artery (2%), and gastroduodenal artery (2%) [5]. Causes of abdominal aneurysms include infection, trauma, surgery, collagen disease, pancreatitis, and MALS [6, 7].

MALS is a disorder caused by compression of the root of the celiac artery by the median arcuate ligament, resulting in decreased blood flow of the celiac artery region [1]. In contrast, the blood supply to the common hepatic artery and the splenic artery originating from the superior mesenteric artery through the pancreatic head arcade arteries is increased. The increased blood flow induces blood dynamic stress to the pancreatic head arcade arteries leading to aneurysms in the pancreatic head area [2]. Approximately 80% of patients with MALS have dilated pancreatic head arcade arteries, whereas 68–74% of patients with an aneurysm of the pancreatic head arcade arteries have MALS [8].

An aneurysm of the pancreaticoduodenal artery ruptures toward the retroperitoneal cavity [9], and the mortality rate of patients with rupture is up to 50–75% [7, 10–12]. Ties et al. reported that 17.6% of ruptured aneurysms of the pancreaticoduodenal artery were smaller than 1 cm in size [13], and all the aneurysms were suitable for treatment. Surgery and interventional radiology using AG are available as treatment options. Because the mortality rate is higher after surgery, interventional radiology is considered to be the treatment of choice [3].

Among the three patients in our study, the physical conditions and the duration between the onset of symptoms and hospital admission were different. In patient 1, hemorrhage extended to the peritoneal cavity, and the patient was admitted to our hospital immediately after the onset of hypotension. In contrast, patients 2 and 3 were admitted over a week after the onset of complaints; in these cases, the hemorrhage was restricted to the retroperitoneal cavity and the volume of hemorrhage was limited. These differences are probably due to diverse patterns of hemorrhage among patients, with different effects on the patients' conditions.

In each patient, the fluid collection was visible around the second portion of the duodenum, and plain CT revealed the fluid collection to be hyper-dense suggesting the presence of blood, which could not be recognized as a hyper-dense area on CE-CT. In a clinical setting, doctors tend to rely on contrast-enhanced CT (CE-CT) to diagnose abdominal

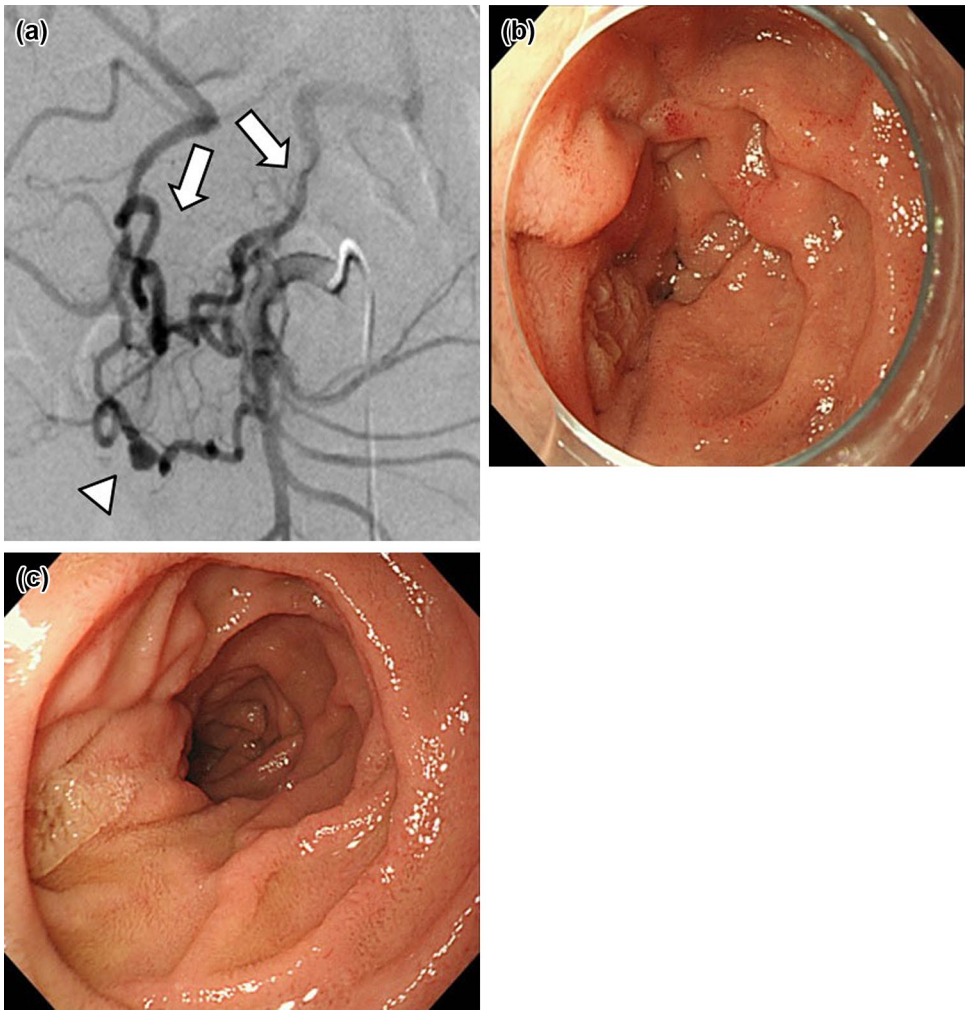

Fig. 3 Case 2. **a** AG through the superior mesenteric artery. An aneurysm of the anterior superior pancreaticoduodenal artery without extravasation (arrow head) is visible. The common hepatic artery and the splenic artery are opacified (arrow). **b** Esophagogastroduodenoscopy (EGD) on 7 days after TAE Impaired extensibility of the second portion of the duodenum. **c** EGD on 20 days after TAE. The extensibility of the second portion of the duodenum improved

disorders, because it provides clearer images with opacified blood vessels than plain CT. However, the slightly hyperdensity of blood could be recognized just on plain CT. That is, we should always check the findings on plain CT in a patient with abdominal fluid collection, and in a patient with acute abdominal pain, plain CT could contribute to diagnosing retroperitoneal or intraperitoneal hemorrhage. If hyper-dense fluid collection around the second portion of the duodenum is recognized, hemorrhage of the aneurysm with MALS could be suspected.

When we see fluid collection suggesting blood around the duodenum or pancreas head, hemorrhage from a pseudocyst with acute pancreatitis, pancreatic arteriovenous malformation (AVM), IgA vasculitis (Henoch–Schoenlein purpura) and tumor should be considered beside hemorrhage due to MASL. These disorders usually have specific findings and the diagnosis is not so hard. Pseudocyst with acute pancreatitis has the episode of acute pancreatitis and the finding of a cyst containing blood. In pancreatic AVM, abdominal angiography shows a racemose intrapancreatic vascular network by arteries and veins [14]. A patient with IgA vasculitis has a specific skin lesions of purpura and significant thickening of the bowel wall accompanied by reduced peristalsis and dilated bowel loops [15], while almost all patients with IgA vasculitis are rarely seen in adults [16].

As a treatment for hemorrhaging aneurysms with MALS, incising the median arcuate ligament could be combined with treatment of the aneurysm to restore blood flow of the celiac artery, thereby decreasing the blood dynamic stress on the pancreatic head arcade arteries [2]. Mere incision of the median arcuate ligament has been reported to be effective for treating aneurysms [17, 18]. Another report described the efficacy of the treatment for MALS with an aneurysm by dilating the stricture of the root of celiac artery using a stent with the treatment of the aneurysm by endovascular embolization [13]. Conversely, some reports did not treat MALS, because no recurring aneurysms were observed after treating the ruptured aneurysm without treatment of MALS [19, 20]. No consensus exists about whether MALS should also be treated in patients with a ruptured aneurysm. In our patients, the median arcuate

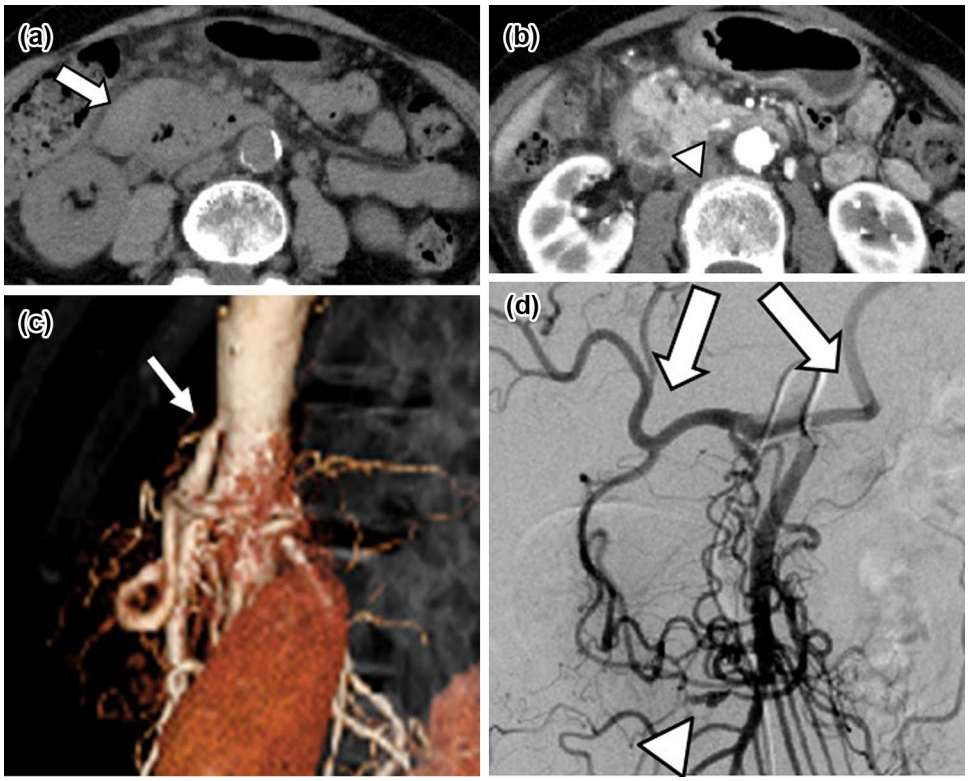

Fig. 4 Case 3. **a** Abdominal plain CT. Hyper-dense fluid is revealed within the fluid collection around the second duodenal portion (arrow). **b** CE-CT. Contrast medium is accumulated within the lesion area (arrow head). **c** CT-AG. A narrowed root of the artery is visible (arrow). **d** AG through the superior mesenteric artery. The common hepatic artery and the splenic artery are opacified (arrows); the aneurysm is visible in a branch of the dorsal pancreatic artery (arrow head)

ligament was not incised, but no recurring aneurysms were observed for up to 6 years after treatment.

Conclusions

Rupture of an aneurysm in patients with MALS is associated with diverse clinical courses depending on the hemorrhage site. In cases with abdominal pain, hyper-dense fluid collection on plain CT could indicate hemorrhage within the abdomen due to rupture of the aneurysm.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Human rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed Consent Informed consent was obtained from all patients for being included in the study.

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