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Gastrointestinal bleeding due to an aneurysm of the pancreaticoduodenal artery in a 7-month-old girl[☆]

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ABSTRACT

Visceral artery aneurysms are uncommon, especially in the pediatric population. We report a rare case of a ruptured saccular aneurysm of the superior pancreaticoduodenal artery (PDA) in a 7-month-old girl presenting with hematemesis. Esophagogastroduodenoscopy showed a constriction of the duodenum and MRI demonstrated a saccular aneurysm of the superior PDA. The infant was taken to the operating room and resection of the ruptured aneurysm was performed. Histologic examination confirmed the diagnosis of an aneurysm, but could not clarify the underlying etiology. This is a rare case report about a female infant presenting with hematemesis due to a ruptured aneurysm of the superior PDA. Ruptured visceral artery aneurysms are a very unusual but serious cause of upper intestinal bleeding, even in infants.

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Visceral artery aneurysms are uncommon, especially in infants. Even in adults aneurysms of the pancreaticoduodenal artery (PDA) are rare, accounting for only 2% of all visceral artery aneurysms [1,2]. Clinical presentation is unspecific and ambiguous [3]. Additionally, delay in diagnosis is common [4]. In children only few cases of visceral aneurysms have been described in the literature. Two girls at the age of 10 and 12 years with ruptured aneurysm of the superior mesenteric artery (SMA) are reported [5,6]. Besides, Esposito et al. presented a case of an 8-year old boy with a small aneurysmatic lesion of the superior PDA causing chronic intestinal bleeding [4]. In this report we describe a rare case of a ruptured saccular aneurysm of the superior PDA in a 7-month-old girl presenting with hematemesis.

1. Case report

A 7-month-old girl presenting with hematemesis was admitted to our hospital. A diagnostic workout was performed. On the

ultrasound study a conspicuous structure between the right kidney and the liver was detected. At first a contrast-enhanced multidetector computer tomography (Fig. 1) was performed, but was unremarkable but for a slight hypervascularization (1.1 × 1.5 cm) near the pancreas. Esophagogastroduodenoscopy showed signs of a previous bleeding and constriction of the duodenum without evidence of active bleeding. Initial hemoglobin level was 6.9 g/dl and dropped to 6.6 g/dl. After blood transfusion hemoglobin-values were stable. A blood coagulation disorder could not be identified. Due to a suspected vascular malformation a contrast-enhanced MR angiography was performed. MR angiography demonstrated a saccular aneurysm of the superior PDA (1.3 × 1.2 × 1.2 cm) with surrounding hematoma (Fig. 2 and Fig. 3). The otherwise healthy infant was taken to the operating room and resection of the ruptured aneurysm was performed (Fig. 4). The initial postoperative course was uneventful. Oral feeding was started on the 5th postoperative day and the infant was on full oral feeding on the 8th postoperative day. Histologic examination confirmed the diagnosis of an aneurysm, but could not clarify the underlying etiology of the aneurysm in this female infant. There was no evidence for fibromuscular dysplasia or Ehlers-Danlos syndrome. Family history was inconclusive for vascular malformations, aneurysms and arteriosclerosis. Two weeks after the surgical procedure the girl refused food intake and lost weight. The diagnostic workout including esophagogastroduodenoscopy showed regular results. Developmental follow-up revealed an adaption disorder after sudden termination of

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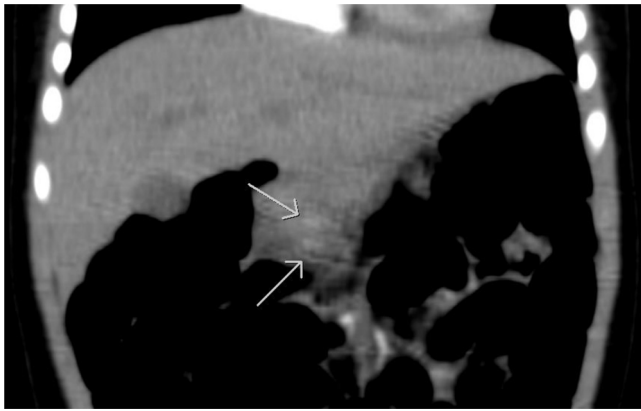


Fig. 1. Contrast-enhanced multidetector computer tomography showing slight hypervascularisation near pancreas.

breast feeding and resultant disruption of the mother-child-relationship.

2. Discussion

Visceral artery aneurysms are rare in adults and even more in children. In adults the majority are of atherosclerotic origin [5]. Other etiologies like fibromuscular dysplasia, Ehlers-Danlos syndrome, pancreatitis or chronic arterial injury from duodenal ulcer are described in the literature [7]. Only few cases of visceral artery aneurysms in children have been reported until today. In one girl the etiology of the aneurysm of the SMA and the underlying cause for rupture is unknown [6]. In another girl histopathological examination identified a connective tissue disease, most likely Ehlers-Danlos syndrome subtype IV [5]. An 8-year-old boy was found to have chronic intestinal bleeding because of a small aneurysm of superior PDA [4]. Visceral artery aneurysms tend to increase in size and carry the risk of rupture with high mortality rate [5,7]. Therefore surgical treatment is recommended even in asymptomatic individuals [8]. The surgical treatment possibilities include aneurysm resection with or without end-to-end anastomosis or venous graft [5]. Ligation without reconstruction is



Fig. 2. Contrast-enhanced MR angiography showing a saccular aneurysm of the superior PDA (1.3 × 1.2 × 1.2 cm) with surrounding hematoma.

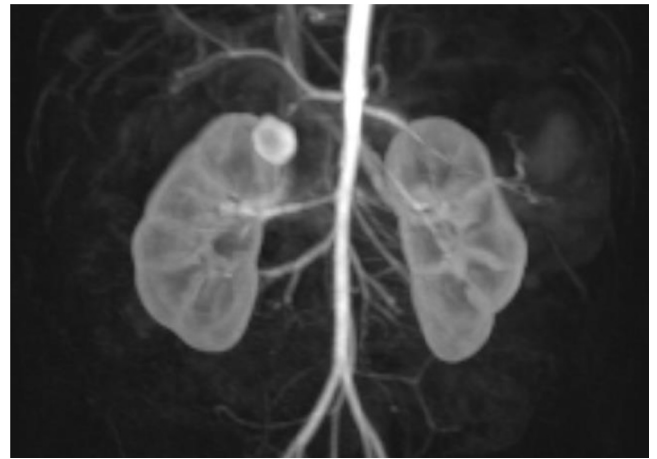


Fig. 3. Contrast-enhanced MR angiography demonstrating a saccular aneurysm of the superior PDA.

preferred in emergency situations for aneurysms of the gastric artery, pancreatic artery and PDA [7]. Additionally, percutaneous US- or CT-guided methods with injection of thrombin in the aneurysm sac are described [5]. Many of the patients present acutely in the emergency ward. Clinical presentation of ruptured aneurysms includes abdominal pain, bleeding into the gastrointestinal tract and hypotension. PDA aneurysms are often associated with pancreatitis leading to periarterial inflammation or vessel erosion from an adjacent pseudocyst [7]. In our case, the underlying etiology of a ruptured PDA aneurysm remains unclear. There was no history of pancreatitis, duodenal ulcer or other inflammatory process.

3. Conclusion

This is a rare case report about a 7-month-old girl presenting with hematemesis without massive hemorrhage or hypotension due to a ruptured aneurysm of the superior PDA. Initial diagnosis

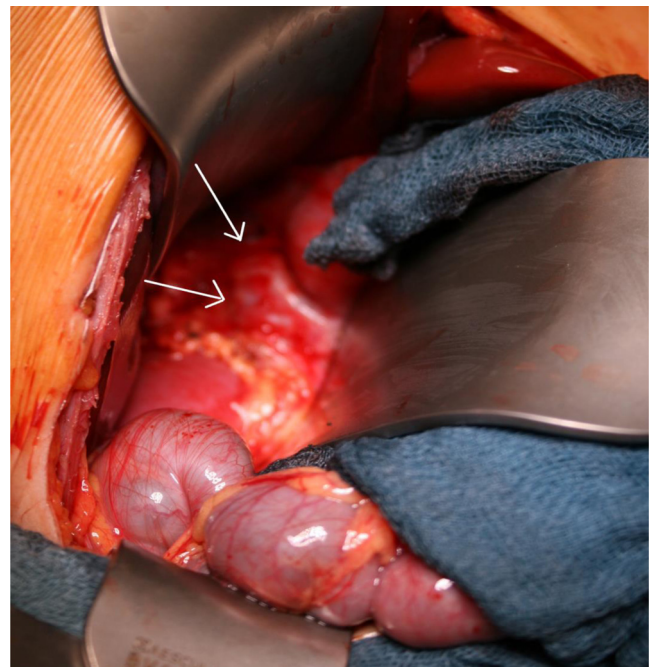


Fig. 4. Intraoperative finding of the aneurysma of the superior PDA during laparotomy.

was difficult, but after appropriate diagnosis in a contrast-enhanced MR angiography operative resection without reconstruction was performed.

In summary, ruptured splanchnic artery aneurysm and especially of the superior PDA is a rare cause for upper intestinal bleeding, even in infants.

Conflicts of interest

The author declares that there are no conflicts of interest.

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