CASE REPORT

Splenic artery pseudoaneurysm due to acute pancreatitis in a 6-year-old boy with acute lymphoblastic leukaemia treated with L-asparaginase

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SUMMARY

Acute pancreatitis is a rare phenomenon in children but its incidence seems to be increasing. In children, it is generally caused due to systemic illness, biliary disease, trauma, idiopathy and side effects of medicines like L-asparaginase. Acute pancreatitis is difficult to diagnose in children since the clinical presentation is highly variable. Complications such as pseudocysts have been reported at rates as high as 25%. Severe cases of pseudocysts may be further complicated by a possible lethal splenic artery pseudoaneurysm. In this case report, we present a rare case of splenic artery pseudoaneurysm due to acute pancreatitis in a 6-year-old boy with acute lymphoblastic leukaemia treated with L-asparaginase. He presented with fever, irritability and pain in his left groin region.

BACKGROUND

Acute pancreatitis is a rare phenomenon in children but the incidence seems to be increasing.^{1 2} In children, it is generally caused due to systemic illness, biliary disease, trauma, idiopathy and side effects of medicines like L-asparaginase.³ Acute pancreatitis is difficult to diagnose in children since the clinical presentation is highly variable but the most common symptoms are abdominal pain, vomiting and irritability.⁴ The disease often has a self-limiting course but about 20% of the cases progress to severe pancreatitis.⁵ It is difficult to identify children who develop severe acute pancreatitis as the scoring system for acute pancreatitis in adults is inappropriate for use in children. The rate for major complications, such as pseudocysts, has been reported as high as 25%.⁶ Severe cases of pseudocysts may be further complicated by a possible lethal splenic artery pseudoaneurysm. Contrast-enhanced CT is the most useful imaging method for evaluating severity and complications but radiation exposure is high, which is especially problematic in children.⁷ In this case report we present a rare case of splenic artery pseudoaneurysm due to acute pancreatitis in a 6-year-old boy.

CASE PRESENTATION

A 6-year-old boy with acute lymphoblastic leukaemia treated with L-asparaginase presented with fever, irritability and pain in his left groin region. Blood tests revealed a high C reactive protein (233 mg/L), leucocytosis ($15\ 610^9/\text{L}$), thrombocytosis ($425\ 10^9/\text{L}$) and a highly elevated p-alanin-transaminase (72 U/L). Neither pancreatic amylase nor lipase was measured at admission.

Figure 1 A 6-year-old boy with acute lymphoblastic leukaemia treated with L-asparaginase presented with fever, irritability and pain in his left groin region. Acute bleeding was suspected. Transversal T2 image (A) showing localised pancreatitis in the cauda of the pancreas (white arrow) and a splenic artery pseudoaneurysm (white arrowhead) was suspected. CT angiography with arterial phase (B) and venous phase (C) confirmed the diagnosis and also identified a possible fistula to the left colon flexure with extravasation of contrast intraluminal in the descending colon (not shown).

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At admission his only symptoms were fever and pain in the left groin region. Physical examination did not reveal any abnormal findings. Myositis or osteitis was suspected and treatment with broad-spectrum antibiotic (meropenem) was initiated. In order to establish the suspected diagnosis, a bone scintigraphy was performed but the findings were normal. After 1 week of treatment the patient improved clinically and biochemically. However, 2 weeks after admission the patient developed diffuse abdominal pain, fever and melena. Haemoglobin levels fell from 7.9 to 6.4 mmol/L and CRP increased from 25 to 70 mg/L. Bleeding from the gastrointestinal tract was suspected. The patient was treated with a proton pump inhibitor (esomeprazol) while gastroscopy and colonoscopy was performed. The endoscopies were without any abnormal findings. In order to identify the source of the bleeding, an MRI of the abdomen was carried out.

INVESTIGATIONS

The MRI revealed localised pancreatitis in the cauda of the pancreas and a splenic artery pseudoaneurysm was suspected. CT angiography confirmed the diagnosis and also identified a possible fistula to the left colon flexure with ongoing bleeding (figure 1).

TREATMENT

Embolisation of the pseudoaneurysm with coils was successfully performed (figure 2). Antibiotic treatment was continued until 2 weeks after embolisation.

Figure 2 A selective angiography of the splenic artery was performed (A) and the exact location of the splenic pseudoaneurysm was confirmed (white arrow). Embolisation of the splenic artery (B) was successfully performed with coils (white arrowhead indicating before and after embolisation) and the bleeding stopped.

OUTCOME AND FOLLOW-UP

One month later, the patient returned to the emergency department with the same symptoms and rebleeding was suspected. However, an acute CT angiography was normal with no signs of ongoing bleeding. The patient was treated conservatively in the intensive care unit and later discharged without any further treatment.

Four months after the first signs of bleeding, the patient had another acute bleeding episode with melena and signs of cardiovascular shock. A new acute CT angiography showed reperfusion of the splenic artery pseudoaneurysm and ongoing bleeding with extravasation of contrast intraluminal in the descending colon (figure 3). Therefore, a fistula to the left colon flexura was once again suspected. Embolisation was once again attempted, but the bleeding could not be stopped. Instead an exploratory laparotomy was performed resulting in splenectomy, distal pancreatic resection and resection of colon transversum.

DISCUSSION

At admission myositis was suspected because of fever and pain in the groin region. A diagnosis of infectious pathogens like shigellose was less likely since diarrhoea was absent. Inflammatory bowel disease or bleeding from the upper gastrointestinal canal was less likely since gastroscopy and colonoscopy were normal. The final diagnosis of acute pancreatitis was not suspected till the MRI gave the diagnosis 2 weeks after admission.

Since the incidence of acute pancreatitis is increasing in children, it is important to consider this diagnosis, especially in

Figure 3 Rebleeding was suspected and a CT angiography with arterial phase (A) and venous phase (B) confirmed rebleeding in the splenic artery pseudoaneurysm and also identified a fistula to the left colon flexure with massive bleeding (not shown). Notice the rapid filling of the pseudoaneurysm with contrast (white arrow) indicating severe ongoing bleeding. The images show reduced blood supply to the spleen and coils in the splenic artery (white arrowhead).

children with systemic illness or those undergoing treatment with L-asparaginase. In these patients, measurement of amylase or lipase should be performed routinely as part of the diagnostics.

In the literature there is an ongoing discussion regarding the use of CT scans in children because of the risk of radiation exposure, which could potentially result in a life-threatening cancer later in life.^{8–12} Here, MRI was chosen as the diagnostic imaging modality to localise the bleeding source and to avoid radiation exposure. This can delay the diagnosis and treatment in a potentially lethal situation and even miss lesions like extravasation. Even though radiation exposure is relatively high for CT angiography, it should always be considered as the initial imaging modality when endoscopy is negative.¹³

In the majority of cases, acute pancreatitis in children is self-limiting; however, because of the high complication rate a scoring system to detect children with acute severe pancreatitis is needed. It seems that the CT severity index (Balthazar score), which is based on the appearance of the pancreas and the extent of necrosis, is the best to predict severe pancreatitis.¹⁴

There is no clear evidence on how splenic artery pseudoaneurysms in children should be treated. If the patient is unstable, surgery should be performed as in adults. However, if the patient is stable, embolisation with coils or endovascular stenting is an alternative treatment option. Puri *et al* reported a 5-year-old boy with splenic artery pseudoaneurysm treated with an endovascular stent graft.¹⁵ They discussed the risk of stent migration, thrombotic occlusion and potential infection. In addition, the small-sized paediatric vessels make it challenging to

place stents. Owing to the risk of stent migration and the anatomic difficulties, this patient was treated with endovascular coils, which has been demonstrated to be a minimally invasive treatment with few complications. In both techniques, close follow-up is needed to avoid episodes of rebleeding.¹⁶

Competing interests None.

Patient consent Obtained.

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Learning points

- Acute pancreatitis is rare in children, but it should be suspected in children with a combination of systemic illness and abdominal pain.
- Acute pancreatitis is a common side effect in children with acute lymphoblastic leukaemia being treated with L-asparaginase.
- In children with gastrointestinal bleeding and no findings on upper endoscopy and colonoscopy, CT angiography is the first choice of imaging. In children suspected of Meckel's diverticulum, laparoscopy should be considered.

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