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Superior mesenteric artery thrombosis in a child with chronic abdominal pain

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ABSTRACT

We present a case of a 13 year old boy with global developmental delay, due to leukoencephalopathy, recent onset of epilepsy and faltering growth. There was a background of chronic abdominal pain, considered to be gastroesophageal reflux disease. He presented with an acute exacerbation of his abdominal pain, abdominal distention and shock. Ultimately a superior mesenteric artery thrombosis was diagnosed and treated. This report focusses on the clinical features of this case along with the diagnostic approach. It illustrates this serious and rare but treatable cause of chronic abdominal pain which can be suspected by careful history taking.

1. Case report

A 13 year old male presented acutely to the emergency department with short history of fever, episodes of crying and green bilious aspiration from his gastro jejunal (G-J) tube. He looked unwell with shallow respirations and reduced respiratory rate of 17 breaths per minute. His oxygen saturation in air was 88%, his chest was clear. He had a sinus tachycardia of 230 beats per minute and a capillary refill time of greater than 3 seconds with cold peripheries. His blood pressure was 89/59 (66). The abdomen was distended. His eyes opened to voice and he responded to pain and there was no rash.

Initially he had a raised lactate (9.1mmol/l), a mild mixed metabolic and respiratory acidosis (pH 7.3; PCO₂ 71 mmHg; bicarbonate 25mmol/ l), evidence of acute kidney injury (urea 19mmol/l; creatinine 101micromol/l; K⁺ 3.8mmol/l) and a raised white cell count (31×10^9 / L) with neutrophilia (28×10^9 /L): platelets and haemoglobin were normal with a low CRP (<1.0mg/l). His electrolytes, liver function and clotting screen were all normal; prothrombin time 18.7 s (12.7–16.1 normal range), INR 1.5 (0.8–1.2), APTT 30.4seconds (26.0–37.0), fibrinogen 3.4g/dl (1.5–4.5).

After initial resuscitation with oxygen, 60ml/kg of intravenous 0.9% saline and commencement of broad-spectrum antibiotics, he was transferred to a Paediatric Intensive Care Unit (PICU) with a provisional diagnosis of sepsis. Further support with intubation or inotropes was not needed at this stage. As he started to respond to resuscitative treatment,

it became more apparent that his abdomen was distended and also tender to examination.

2. Clinical progress

After a surgical review, an abdominal CT with contrast was instigated. This showed small-calibre abdominal aorta with narrow origin of superior mesenteric artery (SMA) containing probable non-occlusive clot, with reduced mid-gut perfusion denoting bowel ischemia (Fig. 1 and Fig. 2). The patient was taken to the operating theatre (OT) for exploratory laparotomy. The surgeon in OT asked for interventional radiology because they were able to palpate clot in the SMA. The interventional radiological procedure showed (conventional angiogram of abdomen and pelvis, with selective SMA angiogram) the clot was subtle and only visible after pixel shift and contrast edge enhancement. Following heparinisation with a dose of 100 IU/kg, mechanical thrombectomy was performed using a Mustang balloon catheter (size 4×40 mm and 6×40 mm). This mechanical thrombectomy was performed via endovascular approach and not direct surgical approach. Post angioplasty imaging revealed SMA clearance with no evidence of residual obstruction. (Fig. 3). Operative inspection confirmed improved perfusion of the small bowel.

Planned re-evaluation of bowel viability at a second laparotomy, 48 hours later, revealed 137 cm of demarcated ischaemic damage of the small bowel which required resection. Primary anastomosis was not

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Fig. 1. Computed Tomography (CT) with contrast: Sagittal view, contrast enhanced abdominal CT showing filling defect within the origin of proximal superior mesenteric artery (red circle) *(images enhanced for contrast and maximum intensity projection)*. Coeliac axis emerges above and laterally, not in the same plane of view. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)



Fig. 2. Angiography: Lateral projection from the biplane aortogram performed by pump injection, showing relative hypodensity (red circle) of the origin of the superior mesenteric artery corresponding to the CT findings. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

possible and ileostomy and de-functioning distal stoma was created. The post-operative course was complicated by stoma leakage with ileostomy stricture requiring further operative reviews and ultrasound guided drainage of an intra-abdominal abscess. Nutrition was maintained



Fig. 3. AP view of the initial SMA balloon angioplasty with the Boston Scientific Mustang $4 \text{ mm} \times 40 \text{ mm}$ balloon showing tapering of the proximal aspect of the balloon before inflation at nominal pressure, denoting thrombus presence.

parenterally in the acute care stages alongside antibiotic therapy and attention to fluid maintenance. He has now recovered and a reanastomosis is planned for the near future.

3. Background history

This child had an underlying leukoencephalopathy diagnosed in infancy on MRI scan and recent onset of generalised epilepsy. A gastro jejunal (G-J) feeding tube had been instituted from 6 months of age because of poor feeding secondary to the neurological condition, his nutritional status and an unsafe swallow. He had been investigated a few years prior to this acute deterioration for chronic abdominal symptoms and faltering growth. Mother gave a two year history of the child's discomfort during feeding and post prandial crying lasting a few hours after feeding. An abdominal ultrasound study, serum CRP, ESR, full blood count, IgE, coeliac screen, thyroid function, liver and renal profile were all unremarkable. The symptoms were attributed to likely gastroesophageal reflux disease and he was treated with esomeprazole. There was incomplete resolution of symptoms.

His regular medication at the time of acute presentation was levetiracetam for his epilepsy and esomeprazole for the abdominal pain. There was no family history of thrombotic illness, connective tissue disease or other inflammatory condition. His recent outpatient weight at 13 years old was 11.3 kg (Z-score; -12.04) and length 92cm (Z-score; -8.59).

4. Discussion

We describe a case of SMA thrombosis in a patient with developmental delay and faltering growth who presented to the emergency department with abdominal pain and bilious aspiration from G-J tube. The abdominal and feeding difficulties had been present for over two years, culminating as an acute presentation at the time of critical perfusion to the small bowel.

SMA occlusion occurs in adults with atherosclerotic arterial disease as part of a generalised arteriopathic picture, with an estimated incidence of 4.3–10 per 10,000 population; it has an associated high mortality [1]. Chronic symptoms of 'abdominal angina' is caused by ischemic pain in the small bowel when the oxygen demand increases after enteral activity after feeding. Thus, it presents as non-specific, colicky, central abdominal pain, related to meals. Avoidance of eating can lead to faltering growth and weight loss. These chronic features may precede a sudden ischaemic event, with acute onset severe central abdominal pain. The initial physical signs of bowel ischemia are not always indicative of the seriousness of the underlying condition [1]. Plain X-ray imaging and ultrasound scan are not diagnostic [2]. The adult patient presents with a metabolic acidosis, disproportionate pain and, if suspected, the diagnosis is confirmed with high resolution CT angiography [3].

In 1990 the first case of SMA thrombosis in children was described in a 16 year old female [4]. This was associated with a perforated appendix and intra-abdominal sepsis. Fibromuscular dysplasia appears to be a cause of the thrombosis in children who presented at aged 4 and 5 years including one fatal case [5,6]. A more recent case in a 3 year old child had acute multi-visceral thrombosis and ischemia including an SMA thrombosis [7]. Traumatic cause of mesenteric arterial interruption and resultant ischemia occurs in non-accidental injury and other causes of severe abdominal trauma in young children [8].

This patient's symptoms had been managed as possible gastrooesophageal reflux disease, a common finding in children with neurodevelopmental problems. The lack of response to treatment is an important pointer towards consideration of this rare underlying cause. On revisiting the history with his mother, he had a clear story of pain during feeding and post prandial crying. SMA thrombosis is a rare cause of recurrent abdominal pain in children but it is worthwhile being wary of this pathology as the diagnosis may discovered by a carefully taken history. The ideal would be for early recognition, before the acute presentation with gut ischemia and need for gut resection.

Patient consent

Consent for publication has been obtained from the family.

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Authorship

All authors attest that they meet the current ICMJE criteria for authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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