



Adhesive subacute small bowel obstruction presenting with gross rectal bleeding

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ABSTRACT

The pathogenesis of massive lower gastrointestinal bleed (LGIB) due to angio-dysplasia and ulceration is rare and not well understood. We discuss an unusual presentation of a child with painless massive LGIB arising from chronic intestinal adhesions causing subacute small bowel obstruction and angio-dysplastic ulceration but without any clinical or radiological evidence of bowel obstruction.

1. Case presentation

An 11 years and 10 months old girl presented to the emergency department (ED) with intermittent episodes of per rectal (PR) bleed for 2 days. On the day of presentation, she was found dizzy and lethargic at home, lying in a pool of blood. She was immediately brought to the toilet wherein she vomited her dinner once with no hematemesis and had another episode of PR bleed. Other than reduced appetite and mild lethargy, there was no prior associated fever, abdominal pain, change in bowel pattern, vomiting or overt gastrointestinal or vaginal bleeding. She denies history of consuming any over the counter or traditional medications. Her last menstrual period was 3 weeks ago with regular flow. Her past history was significant for neonatal ileal atresia. A laparotomy performed at day 3 of life showed a single ileal atresia at the distal ileum approximately 20cm from the ileo-caecal valve. The proximal bowel was dilated and a 10cm segment of small bowel was resected with end to end anastomosis. The appendix was removed and there was no other pathology apparent in the rest of the bowel. She recovered well from this procedure and had remained well until this admission. She had a right congenital auricular agenesis, treated conservatively. Normal left ear and hearing. Her speech, school performance and growth were normal.

On arrival to ED, she appeared lethargic but was lucid and well oriented. Her Glasgow Coma Scale was 15/15. She was hypotensive (BP 68/44 mmHg), tachycardic (HR 111 bpm), afebrile and pale. No jaundice. Pulse volume was reduced, capillary refill time 2–3 seconds, cool

peripheries. Weight 38kg (50th centile) and height 153cm (75th centile). Abdominal examination was normal with no abdominal distension, guarding, tenderness or mass felt. Bowel sounds normal. Rectal examination revealed normal sphincteric tone with minimal blood clots and no evidence of a rectal mass. Rest of her systemic review was unremarkable.

Investigations: haemoglobin 6.9g/dL, platelet count $296 \times 10^9/L$, white cell count $16.2 \times 10^9/L$ (neutrophils 76%, lymphocytes 19%), reduced serum fibrinogen of 1.32g/L (2–4). Prothrombin time, activated partial thromboplastin time, INR and liver profile were normal. Stool culture negative. Supine abdominal radiograph 2 views showed no obstruction or sub phrenic pathology visualised. Ultrasound abdomen was not indicative of intussusception.

Her clinical examination and initial investigations suggested a non-surgical pathology. The diagnosis of a bleeding Meckel's Diverticulum was excluded in view of the findings in the previous laparotomy at birth. She was treated for infective enterocolitis, commenced intravenous cefoperazone/sulbactam and received two pints of packed cells in the first 12 hours.

She continued to have significant episodes of rectal bleeding associated with intermittent hypotension and tachycardia, requiring repeated packed cells transfusions. Since admission, her estimated total rectal blood loss was 1700mls and total volume of packed cells transfused was 2100mls. In view of her large episodic rectal bleed, a surgical pathology was suspected.

An urgent OGDS with visualization up to the second part of

Abbreviations: PR, Per rectal; GIB, Gastrointestinal bleeding; LGIB, Lower gastrointestinal bleeding; OGDS, Oesophagogastroduodenoscopy; OGIB, Obscure gastrointestinal bleeding.

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Fig. 1. Dilated segment of small bowel with dark blood within (D).



Fig. 2. Gross specimen showing the ulcer (arrows).

duodenum performed 24 hours after admission was normal. A colonoscopy was deferred as the child was too ill for a bowel preparation. As the source of bleed was suspected to arise from the small bowel, a CT abdomen followed by an abdominal CT angiogram was performed which revealed no obstruction or vascular pathology.

In view of ongoing PR bleeding and the requirement for repeat blood transfusions, an urgent exploratory laparotomy was performed. A colonoscopy done just prior to the laparotomy revealed fresh blood clots throughout the colon and distal 5cm of the terminal ileum. There were no bleeding points and the colonic mucosa and distal terminal ileal mucosa were healthy and normal. The laparotomy revealed gross adhesions of the small bowel, with subacute obstruction of the terminal ileum about 10cm from the ileo-caecal valve, at the site of the previous anastomosis. After adhesiolysis, it was noted that there was a segmental dilatation at this area, with a large amount of dark blood within (Fig. 1). A 10cm segment of this bulbous small bowel was resected, the whole small bowel was decompressed by suctioning and observed for bleeding. No further bleeding was found from the rest of the small bowel. An end to end anastomosis was performed. Gross assessment of the specimen after surgery showed an ulcer in the dilated portion of the small bowel, very likely to be the source of the bleeding (Fig. 2).

No further rectal bleed was observed post-operatively. She made an

Table 1

Causes of lower gastrointestinal bleeding based on age [6].

Infants	2–5 years	Older
Non-specific colitis	Polyps	Anal fissure
Anal fissure	Anal fissure	Infectious
Milk allergy	Infectious enterocolitis	Enterocolitis
Duplication of bowel	Intussusception	Polyps
Volvulus	Meckel's diverticulum	Inflammatory bowel disease
Hirschsprung's disease	Schonlein purpura	Lymphonodular hyperplasia
Necrotizing enterocolitis	Hemolytic-uremic syndrome	Henoch-Schonlein purpura
Bleeding diathesis	Lymphonodular hyperplasia	Angiodysplasia
	Angiodysplasia	Hemolytic-uremic syndrome
		Bleeding diathesis

unremarkable recovery and was discharged at day 6 post-operatively.

2. Discussion

Gastrointestinal bleeding (GIB) is a common condition in children and can occur in any area of the gastrointestinal tract, from the mouth to the anus. Fortunately, mortality for acute gastrointestinal bleeding (AGIB) is low in the paediatric population [1]. The ligament of Treitz is used as the anatomical landmark to differentiate between Upper Gastrointestinal Bleeding (UGIB) and LGIB. Any bleed that originates proximal to the ligament of Treitz and, in practice, from the oesophagus, stomach and duodenum is classified as UGIB. LGIB in turn is considered if bleeding occurs distal to the ligament of Treitz [2]. Rectal bleeding was identified as the chief complaint in 0.3% of all visits presenting to a busy urban emergency department over a 10-month period. Only 4% of affected patients manifested potentially life-threatening conditions [3, 4]. An appropriate approach to investigate the causes of LGIB is to classify it according to the child's age, general appearance (ill or well), bleeding rate and stool characteristics [1,5,6]. (Table 1)

A detailed history and physical examination should be conducted and always include anal inspection and a digital rectal examination. Tachycardia, pallor, or hypotension suggests significant intravascular depletion that necessitates rapid volume resuscitation and prompt, aggressive diagnostic measures [7].

Our patient presented with an acute painless LGIB with significant intravascular depletion that required immediate packed cell transfusions. A diagnosis of a bleeding Meckel's diverticulum should be entertained in patients with similar presentation. A Meckel's scan was however not done because of the findings from the previous laparotomy at birth. Furthermore, there was no significant history or examination findings as well as radiological evidence to suggest bowel obstruction, severe bleeding from ulcerative colitis, Henoch-Schonlein purpura or haemolytic-uremic syndrome. The volume of PR bleeding and prior good health of our patient would preclude most of the differential diagnosis in Table 1, and make a vascular pathology more likely.

Similar to data from adult patients [8], obscure gastrointestinal bleeding (OGIB) accounts for 5% of all paediatric cases of GIB, including both acute overt and chronic occult types of blood loss. In approximately 75% of OGIB cases, the lesions are detected in the small bowel (mid-GIB) distal to Vater's papilla and reaching as far as the terminal ileum. The source of mid-GIB is related to age, with children showing a greater likelihood of having small intestinal polyps, Meckel's diverticulum, vascular malformations, Crohn's disease, anastomotic ulcers and intestinal duplications [9]. In our case, the overt rectal bleed was considered to be of unknown origin (OGIB) as the CT abdomen, abdominal CT angiogram and endoscopy of the upper and lower gastrointestinal tract did not reveal a source of bleed. The only angiographic sign that is highly specific for acute GIB is extravasation of contrast within the intestinal lumen. This was not apparent in our case. The advantage of

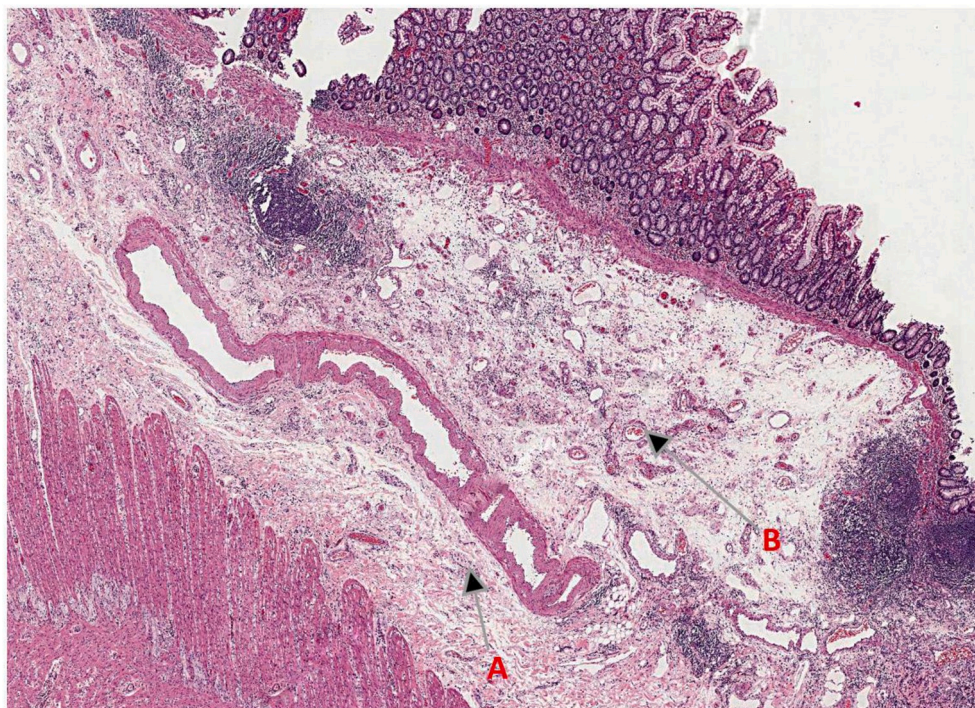


Fig. 3. (x2 magnification) Abnormal vasculature characterised by a dilated vein (A), oedematous submucosa with dilated venules and capillaries (B).

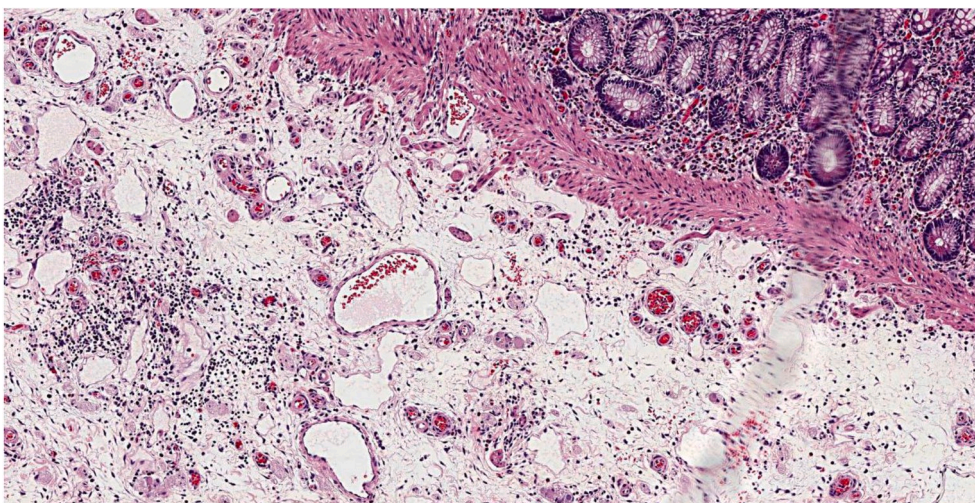


Fig. 4. (x10 magnification) Vascular malformation (angio-dysplasia) showing dilated venules and capillaries in the oedematous, chronically inflamed small bowel submucosa.

angiographic diagnosis of GIB is the ability to perform transcatheter embolization once the bleeding site has been identified.

The exploratory laparotomy we performed, revealed bleeding from an ileal ulcer within a subacute small bowel obstruction due to gross adhesions at the site of the previous anastomosis. If the exploratory laparotomy failed to reveal the source of bleed, an intraoperative enteroscopy, involving insertion of an endoscope through an incision in the mid-small intestine, should be considered. This is usually reserved as a last option to locate the bleeding point.

Intra-abdominal adhesions following abdominal surgery represent a major unsolved problem that can lead to small bowel obstruction [10]. Our case was unusual in that the history, clinical examination and imaging did not reveal any evidence of bowel obstruction. We suspect the adhesions from the previous ileal resection at birth caused the acute on

chronic bowel obstruction. This led to the development of angio-dysplastic vessels lining the endothelium leading finally to ulceration with overt LGIB (Fig. 3 & Fig. 4).

The pathogenesis of angio-dysplasia is not well understood. A proposed theory by Boley et al. [11] is that angio-dysplasia develops due to intermittent, recurrent, chronic low-grade obstruction of submucosal veins at the level of the muscularis propria. Over years, the obstruction results in dilatation and tortuosity of the draining areas (ie, submucosal vessels, venules, and superficial capillaries).

3. Conclusion

Subtle subacute on chronic obstruction appearing years after surgery can be very atypical in its presentation and requires a high index of

suspicion. Angio-dysplasia leading to ulceration and isolated painless rectal bleeding should be considered as a differential diagnosis in patients with previous history of abdominal surgery, despite no evidence of bowel obstruction.

Our case of intestinal vascular angio-dysplasia arising from chronic adhesions which led to intermittent subacute small bowel obstruction and severe LGIB comprises a unique pathologic entity.

Consent

Written informed consent was obtained from the patient's parent for publication in this case report.

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Clinical trial registration

Not applicable.

Declaration of competing interest

No potential conflict of interest relevant to this article was reported.

CRediT authorship contribution statement

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analysis, Supervision, Data curation, Writing - review & editing. **S.S. Tan:** Data curation, Formal analysis, Supervision, Writing - review & editing. **Z.Y. Tan:** Formal analysis, Supervision, Writing - review & editing, Data curation.

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