Ulster Med J 2022;**91(1):**30-31

Case Report

Caecal Diverticulum Causing Catastrophic Gastrointestinal Bleeding in a Child: A Case Report

Mary Patrice Eastwood[1], Irene Milliken[1]

Accepted: 2nd December 2021
Provenance: Externally peer reviewed

Abstract

Solitary caecal diverticulae are rare in children and presentation with massive gastrointestinal (GI) bleeding is seldom reported. We present the case of a 13-year-old boy with a two-year history of abdominal pain and multiple inconclusive investigations presenting with a life threating lower GI bleed. We also review the literature surrounding solitary caecal diverticulae and caecal duplication cysts (CDCs).

Introduction

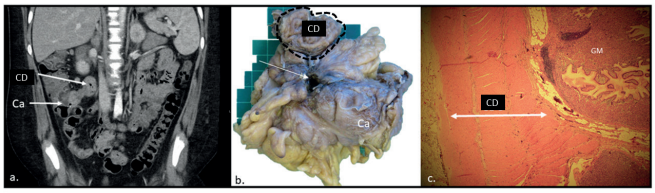
Profuse gastrointestional (GI) bleeding in children, leading to haemodynamic instability is most commonly associated with a Meckel's diverticulum. Herein, we report a rare cause of profuse gastrointestinal bleeding in an older child.

Case Report

A 13-year-old boy (90kg) presented to a referring hospital with a three-day history of bloody diarrhoea, right iliac fossa pain and shortness of breath. On presentation, he was hemodynamically unstable (BP 80 systolic, HR 140 bpm) with a haemoglobin of 34g/L (130-180g/L). His coagulation screen was normal. He was commenced on tranexamic acid, transferred to the paediatric surgical centre and received a massive blood transfusion (6 units). There, a computed tomography angiogram was undertaken which did not demonstrate active bleeding but demonstrated an abnormal tubular structure arising from the ileocaecal valve (Figure 1a).

He reported a two-year history of intermittent abdominal pain with episodes of bloody stool and constipation. In the preceding year, he had had a normal colonoscopy and oesophago-gastro-duodenoscopy. Oesophageal, gastric, duodenal, colonic and terminal ileal biopsies showed no histological abnormalities. His faecal calprotectin was mildly elevated at 90 ug/g and his haemoglobin was 103 g/L at that time. Magnetic resonance enterography had demonstrated terminal ileal thickening with no other areas of inflammation. There had been no concerns on antenatal scans.

Figure 1: a. CT angio showing the relation of the caecum (Ca) to the solitary caecal diverticulum b. Resected specimen demonstrating the caecum (Ca), CD (dashed line) and area of perforation (arrow) were adherent to gonadal vessels c. Histology showing all three layers of diverticulum wall (CD) and hypertrophic gastric mucosa (GM)



In the paediatric surgical centre he underwent a laparotomy which demonstrated an outpouching at his caecum. This outpouching was adherent to his gonadal vessels (Figure 1b). No typical appendix was visualised. The rest of his small bowel and liver were unremarkable. He had a limited right hemicolectomy and primary anastomosis.

Histopathology demonstrated a perforated solitary caecal diverticulum with heterotopic gastric mucosa (HGM) (Figure 1c). No appendix was definitively identified.

At two-month follow-up, he reported to be completely pain-free with no further episodes of GI bleeding.

Discussion

We report a rare case of caecal diverticulum with HGM resulting in life threating GI bleeding in an older child. HGM in a rectal diverticulum causing hematochezia in a 14 year old girl has been reported(1). We identified a single case report of CDC as a cause of rectal bleeding and haemoglobin drop in a 9-month old male (2). This was diagnosed on colonoscopy and ultimately resected(2). Interestingly, they reported benign glandular lining within the cyst wall(2); we were able to confirm HGM in our specimen. Heterotopic gastric mucosa can be found anywhere in the GI tract from mouth to anus. It is typically seen in the oesophagus, duodenum, Meckel's diverticulum or duplication cysts(3).

1 Department of Paediatric Surgery, Royal Belfast Hospital for Sick Children, Belfast, N.Ireland

Royal Belfast Hospital for Sick Children
274 Grosvenor Road
Belfast N.Ireland
BT12 6BA

Corresponding Author Miss Irene Milliken
Email: irene.milliken@belfasttrust.hscni.net
Telephone: 028 905 47666

Profuse GI bleeding resulting in haemodynamic instability does present in children. It is most often described in Meckel's diverticulum which is typically in the absence of abdominal pain, has a quoted incidence of between 1 and 4% and the majority are resected in those under the age of 4 years (4). Vascular malformations arising from the GI tract have been reported to cause rectal bleeding in children occasionally(5). In our patient, GI bleeding from longstanding inflammatory bowel disease had been ruled out on previous investigations. Colorectal carcinoma is seldom seen in children and may cause GI bleeding. It is associated with genetic syndromes such as familial adenomatous polyposis (FAP) and can present with a range of symptoms, unfortunately often late(6). Carcinoid tumours have also been reported to cause massive lower GI bleeding in adults although there are few reports in children(7).

A tubular CDC was also considered but they typically do not communicate with the colonic lumen although it is likely their underlying pathogenesis is shared. Several theories exist as to the origin of enteric duplication cyst. It is suggested that could be secondary to failure in regression of embryonic diverticula, errors of splitting of the notochord or failure in recanalization of the primitive intestine(8).

Enteric duplication cysts are rare in children having an estimated incidence from neonatal autopsies of 1/4500 (8). Colonic duplication cysts account for only 13% of enteric duplication cyst. The majority (80%) of these are cystic, located on the mesenteric side of the bowel and occur in the caecum (8). They contain all layers of colonic wall including muscularis propria(9). They rarely contain gastric mucosa therefore GI bleeding is infrequent, a more common presentation being that of appendicitis(8,9). Similarly, solitary congenital caecal diverticulum are rare, include all layers of the caecal wall and tend to present with pain rather than life-threatening haemorrhage(10).

Conclusion

We report a rare cause of life threating GI bleeding in an older child due to a perforated caecal diverticulum with HGM . Patients with ongoing symptoms and negative investigations may benefit from diagnostic laparoscopy.

Acknowledgements
Special thanks to Dr Caroline Coghlin (pathologist) for her advice and providing specimen and histology images. Furthermore, to Dr Nicholas Clarke (radiology) for providing CT images.

REFERENCES

1. Chen WG, Zhu HT, Yang M, Xu GQ, Chen LH, Chen HT. Large heterotopic gastric mucosa and a concomitant diverticulum in the rectum: Clinical experience and endoscopic management. *World J Gastroenterol*. 2018; 24(30):3462-8.
2. Korotkaya Y, Dunn E, Aybar A, Crino J, Alaish S, Scheimann A. Cecal duplication presenting with hematochezia in an infant. *Clin Pediatr (Phila)*. 2019;58(11–12):1338–40.
3. Yu L, Yang Y, Cui L, Peng L, Sun G. Heterotopic gastric mucosa of the gastrointestinal tract: prevalence, histological features, and clinical characteristics. *Scand J Gastroenterol*. 2014;49(2):138–44.
4. Ruscher KA, Fisher JN, Hughes CD, Neff S, Lerer TJ, Hight DW, *et al*. National trends in the surgical management of Meckel's diverticulum. *J Pediatr Surg*. 2011; 46(5): 893–6.
5. Lee YJ, Hwang JY, Cho YH, Kim YW, Kim TU, Shin DH. A long-segmental vascular malformation in the small bowel presenting with gastrointestinal bleeding in a preschool-aged child. *Iran J Radiol*. 2016;13(1):e29260. doi: 10.5812/iranjradiol.29260.
6. Goldberg J, Furman WL. Management of colorectal carcinoma in children and young adults. *J Pediatr Hematol Oncol*. 2012;34(Suppl. 2):S76–9. doi: 10.1097/MPH.0b013e31824e38c1
7. Dutta G, Panda M. An uncommon cause of lower gastrointestinal bleeding: a case report. *Cases J*. 2008;1(1):235. doi: 10.1186/1757-1626-1-235.
8. Russell KW. Alimentary tract duplications. In: Holcomb JP, Murphy SS, editors. Holcomb and Ashcraft's Pediatric surgery. 7th ed. Elsevier; 2020. Chapter 39. p. 629–40.
9. Lane JS, Sarkar R, Schmit PJ, Chandler CF, Thompson JE. Surgical approach to cecal diverticulitis. *J Am Coll Surg*. 1999;188(6):629–34.
10. Koshy RM, Abusabeib A, Al-Mudares S, Khairat M, Toro A, Di Carlo I. Intraoperative diagnosis of solitary cecal diverticulum not requiring surgery: Is appendectomy indicated? *World J Emerg Surg*. 2016;11(1):1–10.