CASE REPORT

Management of necrotising appendicitis associated with widespread necrotising enterocolitis of the small and large bowel and perforated duodenal ulcer

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Accepted 25 May 2015

SUMMARY

A 7-year-old boy presented in septic shock secondary to appendicitis with generalised peritonitis. Following crystalloid resuscitation, he underwent surgery. Faecopurulent contamination and free air were found. This was secondary to a perforated and gangrenous appendix, multiple large and small bowel segments with perforations, patches of necrosis, interspersed with healthy bowel and segments of questionable viability. There was also a perforated duodenal ulcer. Necrotic segments were resected using a 'clip-and-drop' technique to shorten operative duration and guide resection to preserve bowel length. After six laparotomies and multiple bowel resections, the child was discharged home with an ileostomy that was subsequently reversed. He is currently on a normal diet and pursuing all activities appropriate for his age. Perforated appendicitis can be associated with widespread bowel necrosis and multiple perforations. A conservative damage limitation approach using the 'clip-and-drop' technique and relook laparotomies is useful in the management of extensive bowel necrosis in children.

BACKGROUND

Appendicitis is a common surgical emergency, that in children often presents with perforation due to delay in presentation and/or surgical intervention.^{1 2} We describe a case of necrotising appendicitis associated with multiple large and small bowel segments with perforations, patches of necrosis, interspersed with healthy bowel and segments of questionable viability, and a perforated duodenal ulcer. This led to generalised peritonitis and septic shock necessitating aggressive fluid resuscitation.

An open appendicectomy through a classical incision in the right iliac fossa could have missed some of these findings, especially a perforated duodenal ulcer, with catastrophic consequences. Laparotomy through an appropriately large incision to allow thorough intra-abdominal inspection is obligatory in such complicated cases.

To the best of our knowledge, this is a unique presentation of appendicitis in the paediatric literature with some important learning points for young surgeons and surgical trainees.

CASE PRESENTATION

A 7-year-old boy presented to our emergency department with a 5-day history of abdominal pain and vomiting. He was in septic shock. The

abdominal examination revealed generalised guarding, rigidity and rebound tenderness, consistent with the diagnosis of generalised peritonitis. This was thought to be secondary to perforated appendicitis. Following aggressive crystalloid resuscitation and the start of antibiotic regimen, the patient underwent laparoscopy followed by midline laparotomy.

At surgery, frank pus, faeces and free air were found throughout the abdomen as a result of necrotising appendicitis. On thorough intra-abdominal inspection, multiple perforations of the large and small bowel were found, associated with numerous segments of patchy necrosis of questionable viability spread across both small and large bowel, and a perforated duodenal ulcer (figure 1).

INVESTIGATIONS/DIFFERENTIAL DIAGNOSIS

The differential diagnosis should include pigbel, also called clostridial necrotising enteritis, an often fatal type of food poisoning caused by a β -toxin of *Clostridium perfringens*. Pigbel is almost unique to the highlands of Papua New Guinea, with only one outbreak reported in Europe at the end of World War II. When considering the pathogenesis of pigbel, the epidemiology is important as Papua New Guineans may get pigbel, whereas European residents in Papua New Guinea do not.³

In our case, the microbiology grew *Escherichia coli*, *Streptococcus milleri* and *Pseudomonas*, supporting the clinical finding of appendicitis, with necrotising enterocolitis resulting from sepsis and hypoperfusion.

Since our case focuses on the management of necrotising enterocolitis in a child, with emphasis on a surgical technique that has not been reported in this age group, it may be of benefit in the management of pigbel and is not specific to the primary cause of the enterocolitis.

TREATMENT

The child initially underwent laparoscopy to assess the extent of disease and guide operative approach. Gross faecal contamination, free air and pus dictated a midline laparotomy rather than a traditional transverse incision at McBurney's point. Dense adhesions in the right iliac fossa were dismantled, revealing a gangrenous and perforated appendix in a loculated abscess cavity. The caecum and the ascending colon were almost totally necrotic. There were multiple perforations involving the remainder of the large and small bowel associated with multiple segments of patchy necrosis and areas of

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To cite: Gupta V, Zani A, Jackson P, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-209808

Figure 1 Findings at the first laparotomy.

questionable viability. Thorough examination of the abdominal cavity revealed a perforated duodenal ulcer.

The boy underwent a right hemicolectomy, resection of the necrotic segment of descending colon and multiple resections of frankly necrotic segments of small bowel. The residual segments were tied at either end and placed back into the abdominal cavity in a 'clip-and-drop' method. The duodenal ulcer was dealt with by the insertion of a T-tube into the duodenum through the perforated ulcer. Jackson-Pratt Drains were inserted in the right upper quadrant and left lower quadrant. A triple-lumen central line and peripherally inserted central catheter line were inserted for intravenous fluids, parenteral nutrition (PN) and antibiotics. Neither fascial nor muscle closure was undertaken; the skin was stapled to achieve the abdominal closure.

Second laparotomy (after 48 h)

Findings and intervention: Gross contamination originating from a leak around the T-tube. The tube was removed and the duodenal perforation closed with interrupted sutures and a covering jejunal patch. Two segments of small bowel that were deemed healthy were joined and an ileostomy and gastrostomy fashioned. The skin was stapled to achieve the abdominal closure.

Third laparotomy (after 72 h)

Findings and intervention: Partial breakdown of one small bowel anastomosis and a fresh perforation proximal to the ileostomy with patchy necrosis in the surrounding bowel. The ileostomy was taken down, and ileum resected back to the level of healthy bowel and stapled closed, in a clip-and-drop fashion. The abdominal cavity was lavaged with warm normal saline and closed by skin approximation with staples.

Fourth laparotomy (after 48 h)

Findings and intervention: Small perforation on the antimesenteric aspect of small bowel with several areas of 'impending' perforation. A further 10 cm resection of ileum was undertaken; the necrotic areas were over sewn and healthy ends of small bowel anastomosed. An ileostomy was formed but not matured to the skin due to bowel wall oedema. The fascia in the upper and lower part of the wound was closed and a vacuum-assisted closure device was applied to the residual wound.

Fifth laparotomy (after 48 h)

Findings and intervention: No intra-abdominal collections, healthy but thick-walled bowel prohibiting further maturation of the ileostomy. The fascia was left open in the middle of the wound and the skin was closed with tension sutures around red rubber tubing formed from a Jacques catheter.

Sixth laparotomy (after 4 days)

Findings and intervention: Bile leakage from drain site traced to its origin at the area of the duodenal ulcer repair (despite medical management with an omeprazole infusion). The ulcer was closed around the T-tube and a feeding jejunostomy was created.

Following sixth laparotomy

The child formed a faecal fistula that was successfully managed conservatively.

OUTCOME AND FOLLOW-UP

A month after his initial presentation, the child was discharged with an ileostomy and on full oral diet. Three months later he returned and underwent a straightforward ileostomy closure.

At 26-month follow-up, the patient is on a normal diet and pursuing all activities appropriate for his age.

DISCUSSION

Our case documents extensive small bowel necrosis and perforation along with stress duodenal ulcer perforation in a child with complicated appendicitis. The most likely mechanism of this clinical picture is direct spread of pathogens from the perforated appendix to the remainder of the bowel, effectively causing necrotising enterocolitis.

We have demonstrated that a conservative 'clip-and-drop' approach in managing bowel with extensive segments of perforation and patches of necrosis, interspersed with healthy bowel and segments of questionable viability, can have a positive outcome. Multiple relook laparotomies allowed re-evaluation of disease progression, appropriate resection to control intra-abdominal contamination and conservation of bowel length, preventing short bowel syndrome.

The 'clip-and-drop' technique was first described by Vaughan *et al*⁴ in 1996 for use in infants with multifocal necrotising enterocolitis and later supported by other studies.⁵⁻⁷ In these cases, the multiple necrotic areas are resected, the bowel ends are sealed with titanium clip or staples, and the clipped bowel loops are returned to the abdominal cavity. At a second look laparotomy, the bowel loops can be reassessed and anastomoses can be performed. Its use in trauma laparotomies is well established. This case demonstrates that the technique is transferable as a valuable and effective option in older children with multifocal bowel necrosis. The main advantage of this approach is that it avoids the establishment of short bowel syndrome. Although the outcomes of children with intestinal failure are improving, the morbidity and mortality of long-term PN, bowel lengthening procedures or intestinal transplantation are not negligible and every attempt to preserve native intestine should be made.⁸

Patient's perspective

- ▶ In October 2011 [our son] took poorly with what we was led to believe was a stomach bug but ended up turning our world upside down.
- ▶ From the moment we walked into accident and emergency, we received outstanding care and support from everybody involved. [Our son] and his family are ever so thankful and grateful to Mr Singh and his team for their hard work and for saving [our son's] life.
- ▶ Three and a half years later, [our son] is doing amazingly well and is very proud of his scars. He is always making jokes with his friends saying he got bitten by a shark.

Learning points

- ▶ Complicated appendicitis rarely presents with extensive bowel perforations, patchy necrosis, interspersed areas of bowel of questionable viability, and a perforated stress duodenal ulcer in children.
- ▶ Laparotomy through a large incision and thorough intra-abdominal inspection is a must in such complicated cases.
- ▶ Widespread bowel necrosis in children is optimally managed with a conservative 'clip-and-drop' approach with multiple relook laparotomies.

Acknowledgements The authors would like to acknowledge the patient and his family for allowing us to share this case with our audience and to Queen's Medical Centre, Nottingham, for its support of the paediatric surgery unit.

Contributors VG and SS were involved in the conception and design of the report. All authors were involved in the analysis and discussion of the report and literature, drafting the manuscript, revising it critically and approving the version published.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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