Unexpected outcome (positive or negative) including adverse drug reactions

Ulcerative colitis presenting as acute infectious gastroenteritis with a paralytic ileus

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Summary

A 15-year-old girl who presented with signs of acute infectious gastroenteritis, just as two members of her family is described. As the patient did not improve, a sigmoidoscopy was performed and the diagnosis of ulcerative colitis (UC) was made. Our hypothesis is that an infection triggered the development of UC. Her paralytic ileus was probably triggered by the increased nitric oxide produced in the macrophages and smooth muscles of the inflamed bowel.

BACKGROUND

Inflammatory bowel disease is suspected in children with persistent (≥ 4 weeks) or recurrent (≥ 2 episodes in 6 months) symptoms such as abdominal pain, diarrhoea, rectal bleeding, anorexia, weight loss and arthritis.¹ However, some children have a more fulminant presentation, with severe abdominal pain, bloody diarrhoea, tenesmus, fever, leukocytosis and hypoalbuminaemia. The most common cause of acute diarrhoea is an infection, especially if multiple people in the household have the same symptoms. We present a case of a 15-year-old girl, who initially was suspected of having gastroenteritis but who proved to have an ulcerative colitis (UC), however the diagnosis was not suspected due to its presentation.

CASE PRESENTATION

A 15-year-old female was seen in the emergency department with diarrhoea and vomiting for 7 days. She had lost five kilogram. Both her father and sister were suffering

from the same symptoms. Except for a 3 week period of diarrhoea 3 years previously she had been healthy. She was slightly dehydrated and had a temperature of 38°. Palpation of the abdomen revealed diffuse tenderness. She was orally rehydrated and discharged with a sufficient intake the next day. One day after discharge, she presented herself at the emergency department. She still had diarrhoea, started vomiting again and had severe abdominal pain. Moreover, the complaint of her father and sister were the same except for the abdominal pain. At physical examination bowel sounds were absent. The x-ray abdomen showed no abnormalities. Oral fluids were not tolerated, so she was rehydrated intravenously. The next day she passed 12, unformed, bloody stools, and was incontinent for faeces nocturnally. As she continued to vomit, a central venous catheter was placed and total parental nutrition was started. Due to severe abdominal pain she was completely bedridden.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis in UC includes infectious colitis, parasitic infestations, lymphocytic colitis, collagenous colitis, eosinophilic colitis, medications and pseudomembranous colitis.

INVESTIGATIONS

Stool cultures were negative for *Shigella*, *Salmonella*, *Yersinia*, *Campylobacter*, *Clostridium*, rotavirus, adenovirus and cysts and eggs. The laboratory showed an erythrocyte sedimentation rate of 99 mm, C-reactive protein of 50 mg/l, haemoglobin of 5.9 mmol/l, white cell count of $12.3 \times 10^9/l$, sodium 141 mmol/l, potassium 2.8 mmol/l, albumine of 27 g/l and a faecal calprotectin of 3200 mg/l. Gastro-duodenoscopy showed no abnormalities. Colonoscopy showed a uniformly inflamed mucosa that started at the anorectal verge and extended proximally. Due to the oedema of the mucosa and the fear of perforation the flexura lienalis was not passed. The mucosa showed shallow ulcerations with spontaneous bleeding (figure 1). The biopsies showed inflammation

Figure 1 Coloscopy; shallow ulcerations with spontaneous bleeding.

Figure 2 A small crypt abscess in the deep mucosal layer.

restricted to the mucosal layer with infiltrates consisting primarily of lymphocytes and granulocytes. A small crypt abscess was seen (figure 2). The goblet-cells were depleted, and the crypt architecture showed regularity. Based on the accumulation of these findings the diagnosis of UC was made.

TREATMENT

As oral medication was not tolerated prednisone 2 mg/kg/daily was started intravenously. Due to an unsatisfactory response after 4 days infliximab was started.

OUTCOME AND FOLLOW-UP

A few days after the first dose of infliximab she had no abdominal pain and oral foods were tolerated. She has been on infliximab since and is treated every 8 weeks with 5 mg/kg. Her faecal calprotectin decreased from 3200 mg/l at presentation to 966 mg/l 5 months later and to <20 mg/ml 7 months after the start of infliximab. Three months later a few days before the next dose of infliximab she had some bloody diarrhoea and the faecal calprotectin was 430 mg/ml.

DISCUSSION

Normally UC develops gradually, with a period of diarrhoea, weight loss and bloody diarrhoea. As this patient presented with severe diarrhoea and vomiting and other family members had the same complaints, it was assumed that the child suffered of an acute gastroenteritis. The absence of bowel sounds and the food intolerance in this patient was explained as a paralytic ileus due to the gastroenteritis. These complaints may have been the start of a toxic megacolon (TM), a serious complication of UC. The diagnostic criteria of a TM according to Jalan are radiographic evidence of colonic dilatation. The second criterion is any of the following: fever higher than 38.6°C, heart rate above 120 beats/min, white blood cell count higher than 10.5(10⁹/l) or anaemia. The third criterion is any of the following: dehydration; altered mental status; electrolyte abnormality or hypotension.² Our patient presented with fever, anaemia, electrolyte disturbances and dehydration, the abdomen was tender at palpation and there were no bowel sounds, the x-ray of the abdomen

did not show however colonic distension. The precise mechanism by which colonic dilatation occurs in patients is unknown. A hypothesis to explain TM in UC is that nitric oxide (NO) is generated in the macrophages and smooth muscle cells in the inflamed colon. NO is a known inhibitor of the smooth muscle tone.³ Loss of contractility from the inflamed colon then leads to the accumulation of gas and fluid within the lumen and subsequent colonic dilatation and possibly small bowel dilatation.⁴ Although no infectious pathogens were isolated to explain the diarrhoea of the patient and her family members, numerous infectious agents have been linked to the development and exacerbation of inflammatory bowel disease.⁵ The infectious agent may have triggered the macrophages and smooth muscle cells of the inflamed colon to generate increased amounts of NO. This led to a loss of contractility of the bowel (paralytic ileus). Whether the progression to TM was modified due to loperamide, which the patient received to reduce her vomiting or the analgesics, which she received for her abdominal pain, is unknown. Previously, an unsatisfactory corticosteroid response rate has been described in a meta-analysis by Turner *et al.*⁶ Due to severe vomiting the drugs had to be administered intravenously, limiting the possibilities to either intravenous cyclosporine or infliximab. A randomised comparison of these two drugs has never been performed. Therefore, considering its short-term safety profile, infliximab was started.^{6 7}

Learning points

- ▶ The patient described had an unusual presentation of UC. Our hypothesis is that the patient and her family members, all had infectious diarrhoea, possibly a viral infection. This triggered the UC.
- ▶ Both in the colon and the small bowel the macrophages and smooth muscle cells then generated NO, which resulted in a decreased bowel activity, but no dilatation of the bowel.
- ▶ Hence it is crucial that UC should not be forgotten in clinical decision making in a child with bloody diarrhoea even if all the signs point to infectious diarrhoea.

Competing interests None.

Patient consent Obtained.

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