Chronic campylobacter colitis and uveitis in patient with hypogammaglobulinaemia

Campylobacter jejuni and *coli* are recognised causes of enteritis, and self limiting diarrhoea is a common symptom.¹ Occasionally the disease resembles Crohn's disease and ulcerative colitis. Chronic infection occurs rarely but has been reported in children and patients with immunodeficiency.² We report on a patient with immunodeficiency with chronic diarrhoea in whom *C jejuni* was repeatedly isolated and acute anterior uveitis occurred.

Case report

The patient, a woman, had undergone splenectomy for thrombocytopenia at age 34. In 1974 hypogammaglobulinaemia was diagnosed and treated by gammaglobulin replacement. In 1976 she developed haemolytic anaemia, which was controlled with steroids and two short courses of vinca alkaloids. At that time she started complaining of watery diarrhoea four to six times a day, for which no microbiological cause was found but which improved whenever her steroid dose was increased to control the anaemia. By 1978 she was passing up to 15 watery motions a day, and stool cultures showed *C jejuni* sensitive to erythromycin and tetracycline. She was given erythromycin 500 mg four times a day for three weeks, with no effect on her symptoms or the stool cultures. Her haemolytic anaemia remitted in 1979, and the steroids were gradually withdrawn; her diarrhoea worsened simultaneously. In 1981 she had an episode of acute unilateral uveitis, which was treated with oral steroids, and again her diarrhoea improved.

She was later investigated in hospital several times. *C jejuni* was isolated from her stool on five occasions; two isolates were serotyped and shown to be identical. No giardia, cryptosporidia, or other pathogens were found, and electron microscopy of the stool showed no virus particles. A small-bowel enema was normal apart from the appearance of backwash ileitis. Colonoscopy showed diffuse colitis with multiple small ulcers. Histological examination of the mucosa showed mild inflammation with aphthoid ulceration, neutrophils, no plasma cells, and incipient crypt abscesses, which suggested an infectious cause. Total white cell count was $1660 \times 10^9/l$ (lymphocytes $700 \times 10^9/l$, eosinophils $200 \times 10^9/l$). The urinary concentration of indicans and faecal fat excretion were normal. A Schilling test showed abnormally low vitamin B₁₂ uptake, which was not altered by intrinsic factor. Culture of jejunal biopsy tissue showed moderate growth of commensal bacteria (10^5 organisms/g tissue), as is commonly found in patients with hypogammaglobulinaemia,³ and *C jejuni* (5×10^4 organisms/g tissue). Immunofluorescence techniques using rabbit antibody to *C jejuni* showed organisms on the surface of the colonic mucosa but no submucosal penetration.

She was treated with one 15 day course of tetracycline 500 mg four times daily, five courses of oral erythromycin with doses up to 500 mg four times daily for four weeks, and intravenous gammaglobulin (Sandoglobulin, Sandoz) 9.3 g fortnightly for two months, which did not clear the organism from her stool. On her last admission to hospital in 1982 her diarrhoea was worse and she had lost weight. She was given intravenous erythromycin 600 mg thrice daily for two weeks followed by 500 mg four times daily by mouth for two weeks. Within two days of the start of treatment her bowel frequency had fallen from six to two motions a day. Stool cultures were negative for *C jejuni* and she had no diarrhoea over nine months follow up.

Comment

This case is of interest because uveitis has not been reported in association with *C jejuni* colitis and is not a recognised complication of hypogammaglobulinaemia. Some clinical and radiological features in our patient such as her symptomatic response to steroids were consistent with Crohn's colitis, but the histological findings indicated an infectious cause.⁴ Despite her immunodeficiency and steroid treatment our patient had apparently harboured *C jejuni* infection for about six years without evidence of systemic spread, which confirms that the organism's pathogenicity is mediated by production of enterotoxin.⁵ We suggest that high dose intravenous erythromycin may be effective treatment for chronic watery diarrhoea in patients with hypogammaglobulinaemia.

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Oral acyclovir in eczema herpeticum

Eczema herpeticum is one type of Kaposi's varicelliform eruption. It usually affects patients with atopy who suffer from active eczema, but those whose eczema is in remission may also contract it. People with rare disorders such as pemphigus foliaceus or Darier's disease (keratosis follicularis) are also susceptible. The severity of the infection ranges from localised and mild to widespread and fatal. Patients may die of viraemia, organic complications, or massive secondary bacterial infection, although most recover.

The growth in public awareness of infection with *Herpesvirus hominis* and the increasing incidence of certain varieties of the infection¹ will lead to widened use of the antiherpetic drug acyclovir. I report on two children with eczema herpeticum who were treated with oral acyclovir.

Case reports

Case 1—An 11 year old Sikh boy with lifelong, severe atopic eczema and asthma was admitted to hospital acutely ill with fever, enlarged glands, and widespread impetiginised fissured plaques of eczema, which were most pronounced on his face, neck, and upper trunk. Blood cultures showed contamination with *Staphylococcus epidermidis*, and multiple skin swabs showed heavy growth of *S aureus* and β haemolytic streptococcus group G. He was treated with intramuscular erythromycin, potassium permanganate soaks, and sodium fusidate ointment. With this treatment the crusting slowly cleared and underlying groups of herpetic vesicles could be detected. Because of the degree of sepsis only a scanty quantity of vesicular fluid was obtained for viral culture, but this confirmed a diagnosis of herpes simplex infection. After 48 hours' observation the groups of vesicles seemed to be spreading and he was given oral acyclovir 100 mg every four hours five times daily for five days. This dose was equivalent to 25 mg/kg body weight/day. No

Case 2. Herpetic eruption before (left) and after (right) treatment with acyclovir.

fresh lesions were seen, and his general condition and the lesions improved rapidly.

Case 2—A 12 year old European boy was admitted with fever, considerable herpetiform eruption on his face and neck, and severe angio-oedema of periorbital tissues (figure). The eruption had begun four to five days previously and had spread explosively. He had a history of atopic eczema from infancy, but there had been good spontaneous improvement over the past year or two. No pyogenic organisms were isolated from repeated blood cultures and multiple skin swabs. Virus culture confirmed a diagnosis of herpes simplex infection. The vesicles continued to spread and disseminate, and he was given oral acyclovir 200 mg every four hours five times daily for five days. This dosage was equivalent to 25 mg/kg body weight/day. No further eruption of herpetic lesions occurred (figure), and no evidence of herpetic infection was detected in his eyes. He was also treated with flucloxacillin and sodium fusidate ointment to prevent secondary bacterial infection.

Comment

Both these patients showed a good clinical response to oral treatment with acyclovir, and the infection did not recur during three months of follow up. This treatment caused no discomfort to the patients and had no apparent side effects.² There is not yet an established dosage for children,³ and possibly a higher dosage than was used in these children might have been more beneficial as their renal function was normal and only 20% of acyclovir is absorbed from the gut.⁴

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Cuboidal calcium enterolith causing obstruction and perforation of small intestine

Intestinal strictures are a rare cause of calcium enteroliths, and perforation as a complication has only once been reported.¹ We report on a patient with a cuboidal enterolith; this has never previously been described.

Case report

A 56 year old man was admitted to hospital as an emergency with a three day history of intermittent epigastric pain, which had been severe but had subsequently eased off. He reported having had similar episodes of pain about twice a year for the past few years. His only medical history was of an appendicectomy when he was 11 years old. He had not received any recent medical treatment. On admission his abdomen was slightly distended and tender in the hypogastrium, with increased bowel sounds. Plain radiographs showed distended loops of small bowel and a square opacity, with a lucent centre, adjacent to the left sacroiliac joint (figure). The pain and tenderness resolved within a few hours and he was allowed home.

A week later he was readmitted. On that day the pain had become suddenly more severe and was exacerbated by movement. On examination he was feverish (temperature 37.7°C) and his abdomen rigid. Plain radiographs showed no free gas but some fluid levels in the small bowel. At laparotomy he was found to have peritonitis. His stomach, duodenum, liver, and gall bladder were normal. A large mass was fixed in the left iliac fossa and contained loops of small bowel. The mass was resected and a primary anastomosis performed. Postoperatively he made a good recovery. Subsequent follow up showed him to have gained weight and to be free of pain. Plain abdominal radiography showed no other calcified lesions, and a barium meal and follow through did not detect any abnormalities.

In the resected ileum a 2 cm perforation was found 2.5 cm proximal to an impacted cuboidal hard stone 1.7 × 1.5 × 1.2 cm. Immediately distal to the stone was a fibrous stenosis of the bowel wall. The proximal segment showed

muscular hypertrophy and two ulcerated areas, one of which had perforated. There were no specific features present and in particular no evidence of Crohn's disease, tuberculosis or an ischaemic aetiology.

The stone had hard outer walls composed almost entirely of calcium oxalate. The soft brown core contained some histologically identifiable plant material. Electron probe analysis of the wall confirmed the high calcium content. The patient could not recall having ingested any cuboidal vegetable or fruit.

Comment

Calcium enteroliths are rare and usually develop in the distal small bowel. They are associated with stasis, often related to an intestinal stricture caused by tuberculosis or Crohn's disease.² The more common type of enterolith has a high content of choleic acid and develops in acquired or congenital diverticula.³

Many enteroliths are thought to form around a nucleus, which is usually of plant material, especially fruit skins and stones. Experimental studies have shown that ingested material that swells may cause an obstruction and that oblong objects are slower in their progress through the bowel than round objects.⁴ Fruit pith and skins may produce an ileus at a site of slight stenosis.⁵

Close up of plain abdominal radiograph recorded on admission showing square opacity with lucent centre (↑). Enterolith after removal is superimposed (↑ ↑).

Our patient's long history of intermittent colic suggests that the enterolith had been present at that site for some time and a source of mucosal irritation with subsequent fibrosis. We believe that the most likely course of events was impaction of ingested plant material at a slight ileal stricture without obstruction; formation of a crust of calcium oxalate around this nucleus; excitation of chronic inflammation and fibrosis leading to further narrowing of the segment of the bowel; and finally complete obstruction and perforation. The curious, precise cuboidal shape of the enterolith remains unexplained.

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