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Benign rectal ulceration of unknown origin

An unusual cause of rectal bleeding

Rectal bleeding in children is rarely of serious significance and the most common identifiable causes are superficial anal fissures associated with constipation and benign polyps of the large bowel. Rarer causes of fresh rectal haemorrhage in the young include peptic ulceration of a Meckel diverticulum, intestinal duplication, haemangiomata, and ulcerative colitis. In some patients the origin of the bleeding is never identified and it ceases spontaneously (Gross, 1953). This report describes an unusual cause of repeated rectal bleeding in an infant aged 18 months.

Case report

The patient, the second male child of a Negro Jamaican father and a white mother, presented in August 1973 at 18 months of age with a 6-day history of crying before defaecation and bleeding per rectum. There was no previous history of serious illness or of constipation. Nothing abnormal was found on clinical examination. Haemoglobin was 10·8 g/dl and the blood film hypochromic. Sickle test was negative and stool examination normal. Examination under anaesthetic and sigmoidoscopy showed no evidence of anal fissure or rectal polyp but a small area of ulceration was noted 9 cm from the anal margin. The infant was discharged home but returned after two days with further bleeding. Bleeding and clotting tests were normal and WR was negative. Barium enema examination was also normal.

Rectal bleeding, with and without defaecation, became more frequent in the following weeks and, in October 1973, a limited laparotomy was performed which excluded a Meckel diverticulum as the cause. Rectal biopsy in March 1974 showed normal mucosa adjacent to an area of ulceration and submucosal fibrosis, but there were no specific histological features. The frequency of bleeding increased in spite of treatment with sulphasalazine and rectal steroids. Repeat biopsies in June (Fig. 1) and September 1974 showed increasing submucosal fibrosis. A barium enema in September showed widening of the postrectal space consistent with inflammatory disease of the rectum. An anterior resection of the upper half of the rectum was performed in September 1974 (Fig. 1). In August 1975 the child had had no further symptoms and sigmoidoscopy showed no recurrence of the ulceration in the remaining portion of the rectum or in the colon.

Macroscopic appearance of resected specimen (Fig. 1). The fixed specimen was 6·5 cm in length and 6 cm wide. There was a deep ulcer 2 cm in diameter extending to within 0·5 cm of the distal end. Super-

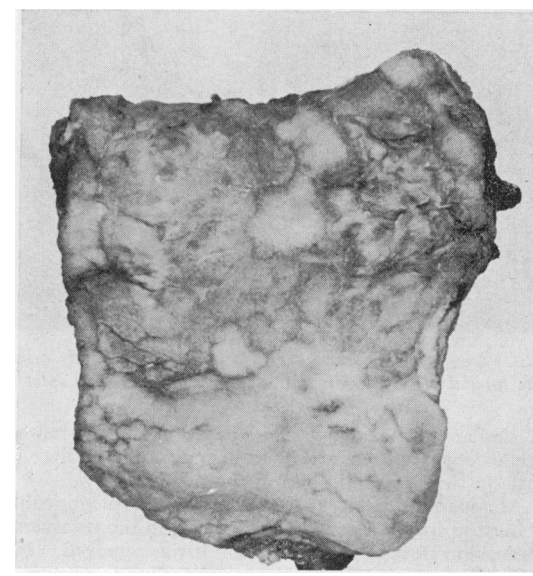

FIG. 1.—*Resected specimen after fixation showing extensive ulceration with islands of mucosa. Distal cut end is at top.*

ficial ulceration with islands of mucosa extended over the distal two-thirds of the specimen.

Microscopy (Fig. 2). Sections from the area of superficial ulceration showed generalized loss of mucosa

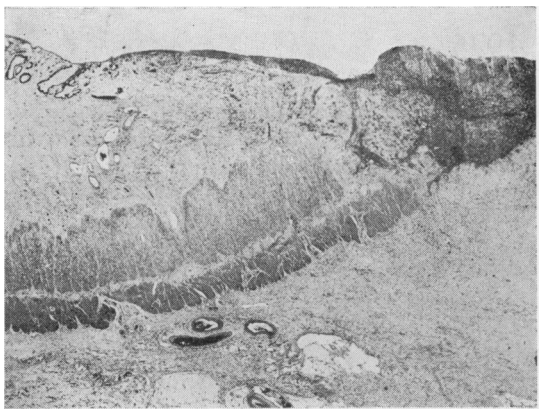

FIG. 2.—*Section of resected rectum through edge of deep ulcer. There is loss of mucosa and destruction of muscularis propria to right side of section.* (*H. and E.* ×7.)

and muscularis mucosae with oedema of the submucosa. The muscularis propria had disappeared in the area of deep ulceration where there was dense fibrosis but only a mild inflammatory cell infiltrate. Iron deposition was absent and there was no abnormality of blood vessels. The mucosa adjacent to the area of ulceration was normal. The microscopical features were therefore nonspecific.

Discussion

This case of nonspecific ulceration of the rectum is presented as a rare cause of rectal bleeding in childhood, although the aetiology of the lesion remains obscure. Stercoral ulceration of the large bowel has been reported in children (Gross, 1953) but there was no history of constipation in this patient. Detailed histological examination showed none of the specific features of Crohn's disease or ulcerative colitis. It is tempting to relate this case to the condition of 'solitary ulcer', or 'colitis cystica profunda,' described in adults (Haskell and Rovner, 1965). However, fibrosis affected the full thickness of the rectal wall whereas in 'solitary ulcers' the muscularis propria usually remains unaffected. Further, in a typical example of 'solitary ulcer' mucosal elements are found within the submucosa (Wayte and Helwig, 1967), and this feature was not seen in the present resected specimen of rectum. The possibility of a bizarre form of repeated rectal trauma cannot be completely excluded but no evidence has been obtained for this diagnosis.

Summary

An unusual progressive form of benign rectal ulceration was found in an infant who presented with repeated episodes of rectal bleeding. The ulceration failed to respond to medical treatment but was cured by anterior resection of the rectum. The aetiology is not known.

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