---Case Report---

SOLITARY ULCER OF THE RECTUM: REPORT OF A CASE AND REVIEW OF THE LITERATURE

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Summary

A 13-year-old girl with Prader-Willi syndrome was admitted to our hospital with an 18-month history of anal bleeding and mucus discharge on defecation. Physical examination revealed obesity, hypogonadism, hypotonia and hypomentia. On digital examination, a nodular mass was palpated on the right wall of the ampulla recti, which was suspected to be carcinoma on a barium enema study. Proctoscopic examination revealed a large, irregular ulceration with white slough at the base, surrounded by the nodular and lumpy mucosa.

The lesion was excised by the abdomino-anal pull-through method. The resected specimen showed a lesion of large, shallow, irregular ulcer, 5.0 x 2.2 cm in size. Microscopic examination revealed obliterated lamina propria by fibroblasts and muscle fibers derived from the muscularis mucosae, and misplaced cystic dilated glands in the submucosa at the margin of the ulcer. The gross and microscopic appearances are identical to those of "solitary ulcer of the rectum" described by Madigan and others, and similar to those of "colitis cystica profunda" described by Goodall and others. According to these findings, this lesion was diagnosed as solitary ulcer of the rectum.

In the present report, the relationship between solitary ulcer of the rectum and colitis cystica profunda was discussed.

Key Words: solitary ulcer of the rectum, colitis cystica profunda, rectal cancer, rectal bleeding, Prader-Willi syndrome.

Introduction

Solitary ulcer of the rectum is very rare. In the present study we report a case of solitary ulcer of the rectum with Prader-Willi syndrome. Twenty other cases of solitary ulcer of the rectum are reviewed and compared with the 11 cases of colitis cystica profunda reported in Japan.

Case

A 13-year-old girl with an 18 month-history of intermittent rectal bleeding was admitted to our surgical department on January 5, 1977. She was the first-born child of a 28-year-old mother. The family history was unremarkable. Pregnancy and delivery were uncomplicated. Weight and height at birth were 2,800 gm and 50 cm respectively. Hypotonia and difficulty in feeding or poor sucking were noticed soon after birth. Psychomotor development was delayed.
At the age of three, she began to thrive and rapidly gained weight. She was admitted to the pediatric department in our hospital at the age of nine and half years for a complete evaluation.

**Physical Examination**

Physical examination revealed marked obesity and short stature. Muscle tone was poor. There was no abnormal finding in the chest, heart or abdomen. Digital (rectal) examination revealed a walnut-size nodular mass on the right anterior wall of the rectum. Obvious rectal prolapse was not noticed.

**Laboratory Findings**

Routine laboratory data were within normal except for a low hemoglobin value. The serologic test for syphilis was negative and tuberculin reaction was positive, 2+ in the first strength. Examination of faeces was positive for occult blood, and was negative for parasite or tubercule bacilli.

**Barium Enema Study**

Barium enema demonstrated irregularity of the right anterior wall in the lower and mid rectum (Fig. 1). There was an irregular filling defect with central shallow barium fleck, suggesting an irregular shaped ulcer. Despite the young age of the patient, the lesion was suspected to be malignant.

**Sigmoidoscopy**

Sigmoidoscopy showed a large, irregular ulceration extending from 2 to 7 cm proximal to anal verge on the right anterior wall of the rectum (Fig. 2). The ulceration was shallow and covered with slough at the base as shown in Fig. 2. This ulcer was surrounded by the nodular and lumpy mucosa, which showed a resemblance to Borrmann II type carcinoma. However, by multiple biopsies from ploypoid and ulcer lesions, it was diagnosed as benign.

**Surgical Treatment and Resected Specimen**

The rectal resection was performed with a tentative diagnosis of colitis cystica profunda or solitary ulcer of the rectum. The operation was made by the pull-through method. Resected specimen showed a large, shallow and irregular ulcer, 5.0 x 2.2 cm in size. The surrounding