Ulcerative colitis in an infant aged 20 months. A case report

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ABSTRACT

Bloody diarrhea is a common problem in early childhood, typically caused by anal fissures, infectious enteritis, allergic proctocolitis, swallowed maternal blood and intussusception. More rarely, it can also be caused by volvulus, coagulopathies, necrotizing enterocolitis, polyps, Meckel diverticulitis and inflammatory bowel disease (IBD).

The incidence of IBD is on the rise in children, even affecting infants. The most common subtypes are Crohn's disease (CD) and ulcerative colitis (UC). While IBD occurrence peaks in the second to third decades of life, paediatric IBD accounts for 7-20% of all cases. Within this age group, the highest rates are seen in the teenage years; however, very early onset IBD can be seen before six years of age. The classic symptoms of CD include abdominal pain, diarrhea and weight loss, while UC is typically associated with bloody diarrhea.

The report describes the case of a 20-month-old boy with bloody diarrhea who was ultimately diagnosed with UC.

Key words: *diarrhea infant, blood, inflammatory bowel diseases, child.*

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INTRODUCTION

Bloody diarrhea represents a challenging diagnostic problem in children. It has been attributed to a broad range of possible causes, including anal fissure, infectious enteritis, allergic proctocolitis, juvenile polyps, Meckel's diverticulum, intussusception, volvulus, coagulopathies, necrotizing enterocolitis, Hirschsprung's disease and inflammatory bowel disease (IBD). The most common subtypes of IBD are Crohn's disease (CD) and ulcerative colitis (UC). The classic symptoms of CD are abdominal pain, diarrhea and weight loss, while UC typically presents with bloody diarrhea. Although the potential diagnosis varies depending on age, intestinal infections play a significant role in all cases. However, in infants, the most likely cause is allergic colitis.

IBD may occur at any age, but is more commonly observed in older children.[1] The Paris classification for IBD stratifies pediatric IBD according to age: a younger category of children diagnosed younger than 10 years of age, and an older category - between 10 and 18 years of age.[2] Approximately 1% of pediatric IBD have been diagnosed in children younger than one year of age, and approximately 15% before six years of age.[3,4] The incidence of pediatric IBD is increasing, particularly in young children.[5-7]

Although the exact etiology of IBD remains unclear, it is thought to be due to a complex interaction between many factors. Apart from environmental factors, defects in the innate and adaptive immune system, microbial dysbiosis and genetic predisposition are all believed to play roles.[8]

CASE REPORT

A 20-month-old boy was referred to the Department of Paediatrics, Allergology and Gastroenterology for bloody diarrhoea. The medical history indicated that pregnancy and post-natal development had proceeded without any abnormalities. The boy had not been hospitalised previously. Within the four weeks preceding hospitalisation, he had experienced as many as seven bowel movements a day, and had loose stools with blood and mucus; otherwise, he reported no other complaints.

Laboratory tests were performed in an outpatient setting: CBC (complete blood count), CRP (C-reactive protein), ESR (erythrocyte sedimentation rate) and stool culture results were all within norms. Outpatient treatment included nifuroxazide and probiotics, and an elimination diet (extensively hydrolyzed casein formula)

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was introduced. Due to lack of improvement, the child was referred to the Department. On admission, the general condition of the patient was good, with no apparent abnormalities. During hospitalisation, the patient performed five to seven bowel movements a day, with bloody stools. Six days later, the boy lost his appetite and had developed a fever; he also passed six to eight mushy stools each day, with < 50% of the stool volume being blood. Some bowel movements occurred at night. No abdominal pain was observed and the child did not demonstrate limited activity. A 600 g loss of body weight was observed within one week.

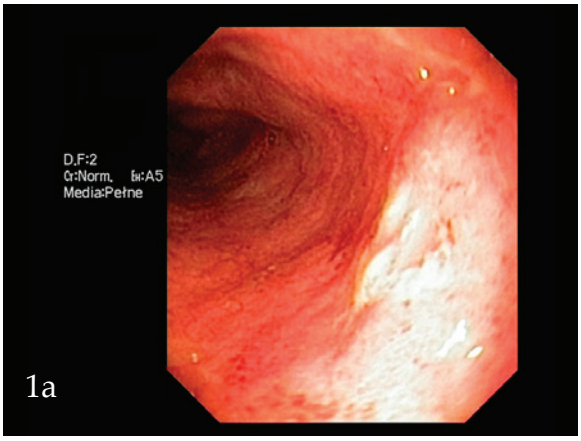
In order to perform a differential diagnosis, microbiological testing was performed. Stool culture did not demonstrate the presence of any viruses (norovirus, adenovirus or rotavirus) or bacteria (*Salmonella, Shigella, Yersinia enterocolitica, Clostridium difficile, Campylobacter jejuni*). Parasitic infestation was also excluded. Allergy testing was also carried out; however, tests for levels of specific IgE with food allergens were negative. An elemental diet was applied for diagnostic purposes.

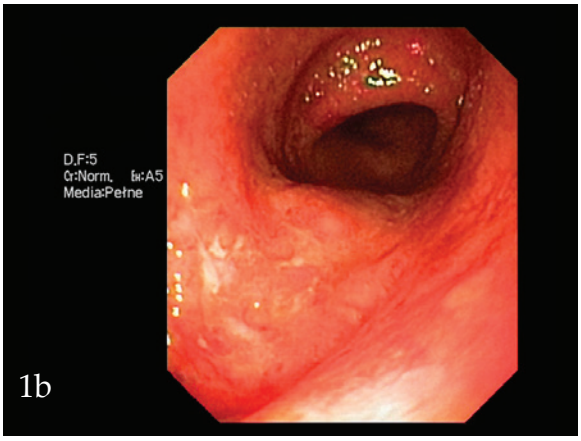
The results of laboratory tests are shown in *Table 1*. The abdominal ultrasound was normal. Endoscopy of the upper and lower gastrointestinal tract was performed. Based on its findings, colitis was confirmed and UC was suspected. Ileocolonoscopy found the patient to have contiguous mucosal inflammation beginning at the rectum and extending proximally to the hepatic flexure, with no macroscopic lesions observable in the terminal ileum, cecum or ascending colon. Diffuse erythema was observed, as were submucosal haemorrhage, friability, granularity, loss of vascular pattern and superficial ulcerations (*Figure 1a, 1b*). Histopathological examination revealed cryptitis, crypt abscesses and severe lymphoplasmatic and neutrophilic infiltrations in the lamina propria of the rectum, sigmoid, descending and transverse colon, and only mild infiltrations in the terminal

TABLE 1. *Laboratory results*

Parameter	Initial values	Follow-up values
Hemoglobin g/dL	10.4	9.2
RBC (10^6/uL)	3.66	3.36
MCV (fL)	82.5	84.2
CBC (10^6/uL)	14,13	17,61
Platelet (10^9/L)	394	279
Albumin (g/dL)	3.1	3,8
Alpha-1-globulins (g/dL)	0.7	-
Gamma-globulins (g/dL)	0.4	-
CRP mg/dL	3,85	0,63
ESR (mm/1sth)	32	20
D-dimer (ng/mL)	2477	905
IgA (g/L)	0,5	-
IgM (g/L)	0,94	-
IgG (g/L)	6,2	-
Na (mmol/L)	139,9	137,3
K (mmol/L)	3,7	4,3
Aspat (U/L)	26	-
Alat (U/L)	11	-
Amylase (U/L)	51	-
TSH (uIU/mL)	0,889	-
FT4 (ng/dL)	1,03	-

FIGURE 1a and FIGURE 1b. *Endoscopic features of UC: diffuse erythema, submucosal haemorrhage, friability, granularity, loss of vascular pattern and superficial ulcerations*





ileum, cecum and ascending colon (*Figure* 2). No macroscopic lesions were seen during upper endoscopy, which was unremarkable, with only mild gastritis observed at histopathology. Disease activity was assessed as 45 points on the PUCAI (Paediatric Ulcerative Colitis Activity Index). The final diagnosis was set as UC.

Treatment included IV methylprednisolone; in addition, ceftriaxone and metronidazole were administered, together with an elimination diet, and intravenous iron formulas were prescribed due to progressive anaemia (HGB 8.6 g/dL). Gradual clinical improvement was observed from week 2 of treatment: the number of bowel movements decreased, the stools no longer contained blood and the body temperature returned to normal. Mesalazine was added. On the day of discharge, the disease activity was assessed to be 30 points on the PUCAI scale (four stools a day, night bowel movement still present, mushy consistency).

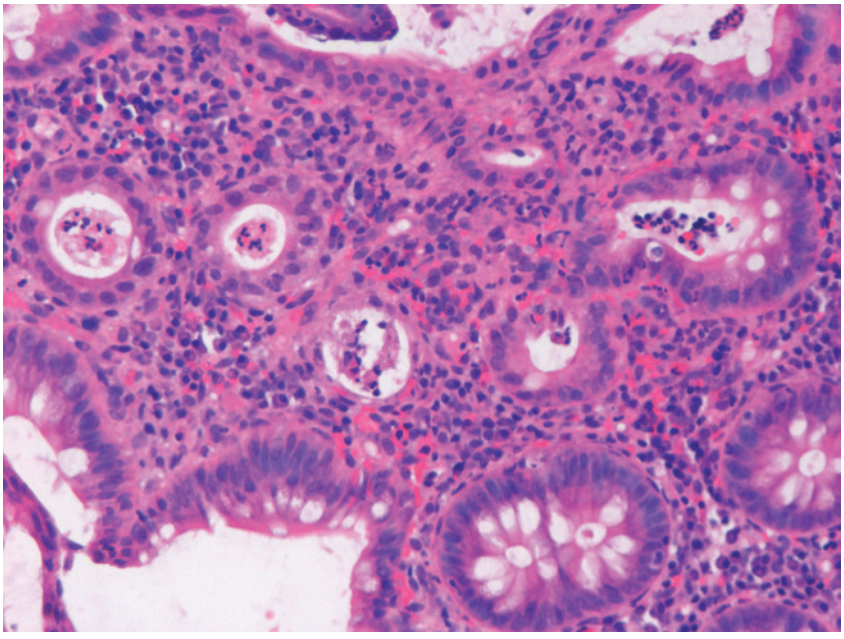
The patient's general condition was good and he was discharged home with a recommendation to continue oral treatment with glucocorticosteroids, mesalazine and to use an elimination diet. Two weeks after discharge, during a follow-up visit at the Outpatient Clinic, the PUCAI scale was assessed to be 10 points, due to bowel movements still occurring at night. The boy felt good, his fever had subsided, his appetite was good, and his body weight had increased. He was passing two bowel movements a day, and the stools had of a normal consistency, with no blood or mucus. A follow-up complete blood count also revealed improvements in red blood cell parameters.

It was recommended to reduce the glucocorticosteroid dose gradually, to continue mesalazine, to expand the range of the diet slowly and attend regular follow-up visits at the Outpatient Clinic. Eight weeks after discharge, the patient was eating without any restrictions and was continuing mesalazine treatment. Currently the child is in clinical remission and is receiving only mesalazine.

DISCUSSION

The main reason for hospitalisation of the patient described in the present study was bloody diarrhoea, which is most commonly caused by the presence of infectious factors. However, both gastrointestinal infection and parasitic infestation were excluded. Another possible common reason is allergic colitis. However, food allergy was also excluded based on the lack of improvement observed following the elimination diet, the

FIGURE 2. *Histology specimens of colon mucosa of the large intestine*



Infiltration of lymphocytes, plasmocytes and granulocytes in the lamina propria, cryptitis and the destruction of individual crypts. Visible mobilization of granulocytes in the vessels. Zoom with the lens 20x.

negative sIgE, no history of allergy (no other allergic symptoms, except bloody diarrhoea; no family history; onset at about 1.5 years old), the negative results of endoscopy with biopsies, the absence of eosinophilic infiltrate in colonic mucosa, and the lack of any apparent worsening after the introduction of a normal diet.

A diagnosis of IBD in infants and young children most commonly includes conditions associated with immunodeficiencies such as chronic granulomatous diseases, Wiskott-Aldrich syndrome, IPEX syndrome, common variable immunodeficiency and IL-10 deficiency.[9-12] In the present case, blood tests were performed to identify immunoglobulin levels, and no lymphopenia or granulopenia was observed; these findings, combined with the patient history, allowed immunodeficiencies to be excluded as a cause. Other, rarer reasons for bloody diarrhoea mentioned above, were excluded.

As pancolitis is an extremely frequent manifestation of IBD in children, being identified in more than 80% of cases, it is difficult to differentiate between UC and CD.[7] UC is characterized by recurring episodes of inflammation of the mucosal layer of the large bowel which always involves the rectum and can extend proximally in a continuous fashion. The diagnosis of UC is based on the results of medical history, physical examination, laboratory tests, imaging studies of the small bowel, upper and lower endoscopy, as well as histological findings. In order to exclude CD, endoscopy of the upper gastrointestinal tract is also performed.[1,13]

In this patient, the diagnosis of UC was supported by typical inflammatory lesions beginning in the rectum and progressing proximally in a contiguous and circumferential fashion. As neither the macroscopic nor microscopic picture of the upper and lower gastrointestinal endoscopies revealed any signs typical of CD, UC was diagnosed in our patient. ANCA play an important role in the pathogenesis of UC, but they were not detected.

Pediatric gastroenterology uses a separate classification for patients experiencing the onset of IBD while below the age of five years: this being a population undergoing intensive development and growth.[14] These children required medical consultations, hospitalisations or surgeries for their underlying disease more rarely than children over 10 years.[6] In contrast, Ledder et al., in children with UC below six years, found that the disease was more aggressive and the children demonstrated significant body weight loss, required a higher supply of immunosuppressive agents and a more frequent need for surgical interventions than older patients.[15]

A differential diagnosis of bloody diarrhoea in the youngest children should include age-typical reasons such as gastrointestinal infections or food allergies, but IBD should also be considered. ■

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