Case Report

Hematochezia in a 6-year-old girl: An unusual cause

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ABSTRACT

Anorectal disorders, anal fissures, and distal polyps are the most common causes of hematochezia in children. However, inflammatory bowel disease (IBD), even though quite rare in children, can present with an isolated per rectal bleeding. Early onset IBD is described in patients younger than 10 years of age; however, these are rare instances of even neonates presenting with IBD. Here, we report the case of a 6-year-old girl who presented with episodes of per rectal bleeding for several months with severe anemia and malnutrition and was also found to have an anteriorly displaced anus. She underwent colonoscopy which revealed features suggestive of ulcerative colitis which was eventually confirmed by biopsy. She was treated with corticosteroids and aminosalicylate, namely, Mesalamine. Later, the patient was treated with Azathioprine which resulted in the complete resolution of symptoms and improvement in nutritional status that was evident on regular follow-ups. IBD in children younger than 10 years of age is a rare entity and requires a strong suspicion for diagnosis.

Key words: Anteriorly displaced anus, Inflammatory bowel disease, Rectal bleeding, Ulcerative colitis

The most common causes of hematochezia include necrotizing enterocolitis in neonates, intussusception in infants, bacterial colitis and rectal or anal polyp, anal fissure in young children, and inflammatory bowel disease (IBD) in children and adolescents. The term IBD accounts for two distinct idiopathic inflammatory conditions of the bowel known as Crohn's disease and ulcerative colitis (UC) [1]. The onset of the illness has a bimodal distribution, around 10–20 years of age, and the subsequent, smaller peak occurs in the geriatric age (50–80 years) [1]. However, there are reports of early-onset IBD [2,3] manifesting even in the 1st year of life, attributed to the increased use of antibiotics at these early ages [2]. Genetic and environmental factors are responsible for the pathogenesis of this disease [1]. Although more prevalent in the West, a rise in incidence is observed in Asian countries as well. Furthermore, the diagnosis of IBD is a challenge in a country like India, where infectious diseases, especially tuberculosis, are prevalent [4].

We report here the case of a young girl, who presented to us within the first decade of life, rather a rare instance, with episodic per rectal bleeding for several months with severe anemia and malnutrition who eventually got diagnosed with early UC. The rationale for reporting this case is that this condition is quite rare in very young children and hence, awareness of this as a possible differential diagnosis in a child with per rectal bleeding would be important.

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CASE REPORT

A 6-year-old female child presented to our hospital, with complaints of frank bleeding per rectum for 4 months. The symptoms were insidious in onset with the passage of about 10 ml fresh blood after each episode of defecation, 1–2 times per day. Stools were normal in color and consistency. There was a history of low-grade fever 3 weeks ago lasting for 2 days, which was treated symptomatically. The mother had noted a few papular skin eruptions involving the trunk and extremities before the development of the bleeding episodes. She did not have abdominal pain or distension, loose stools, or vomiting associated with these symptoms. There was a history of weight loss in the child over the past 3 months in spite of a good appetite. On clinical examination, the child was euthermic and hemodynamically stable. The patient was undernourished with a body mass index of 14.5 and had significant pallor, with platynychia and koilonychia noted over hands and feet (Fig. 1a). Multiple healed, flat, and hyperpigmented lesions were noted over the trunk and extremities (Fig. 1b). Anteriorly displaced anus, with an anal position index (API) of <0.34 (Fig. 2), was detected on perineal examination. The API was calculated using the formula: Fourchette-Anal distance

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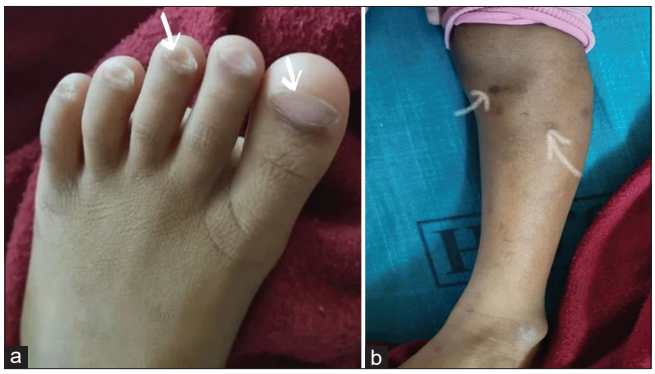

Figure 1: (a) Platynychia and koilonychia of the toe nails and (b) multiple healed hyperpigmented lesions on the extremity

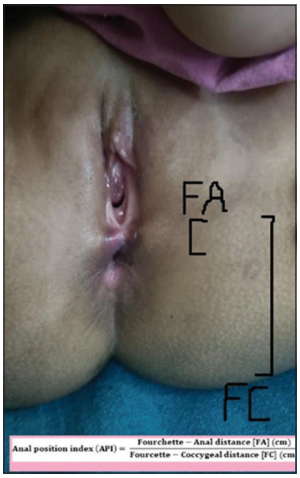

Figure 2: Anteriorly displaced anus. FA: Distance between fourchette and anus; FC: Distance between fourchette and coccyx

(cm)/Fourchette-Coccygeal distance (cm). Systemic examination including per rectal examination was unremarkable. The initial suspicion of a local cause, such as an anal polyp or solitary rectal ulcer, was made, in addition to the anteriorly placed anus also being a possible cause.

The blood investigations revealed severe anemia (hemoglobin 5.2 g%), leukocytosis (23,340 cells/mm^3) with neutrophilic predominance, and thrombocytosis (platelet count 11.38 lakh/mm^3). Peripheral blood smear and iron profile were suggestive of iron deficiency anemia. Stool examination showed plenty of red blood cells, but no inflammatory cells or parasites were found. Inflammatory markers, liver, and renal function tests were normal. HIV serology and Mantoux test were negative. Ultrasound abdomen and pelvis showed multiple non-significant mesenteric lymphadenitis. The gastroenterologist was consulted and the child underwent colonoscopy, which revealed loss of vascularity, friability, granularity, and a few tiny clean-based mucosal ulcers involving the colonic mucosa, suggestive of pan-colitis (Fig. 3).

Histopathology of the mucosal biopsies taken from the ileum, hepatic flexure of the colon, and the rectum showed unremarkable ileal mucosa, hepatic flexure ulcer with moderate focally active colitis along with fragments of colonic mucosa displaying moderate inflammatory cells comprising of lymphocytes, plasma cells, scattered eosinophils, and occasional crypt abscesses, with rectum showing mild colitis with lamina propria displaying

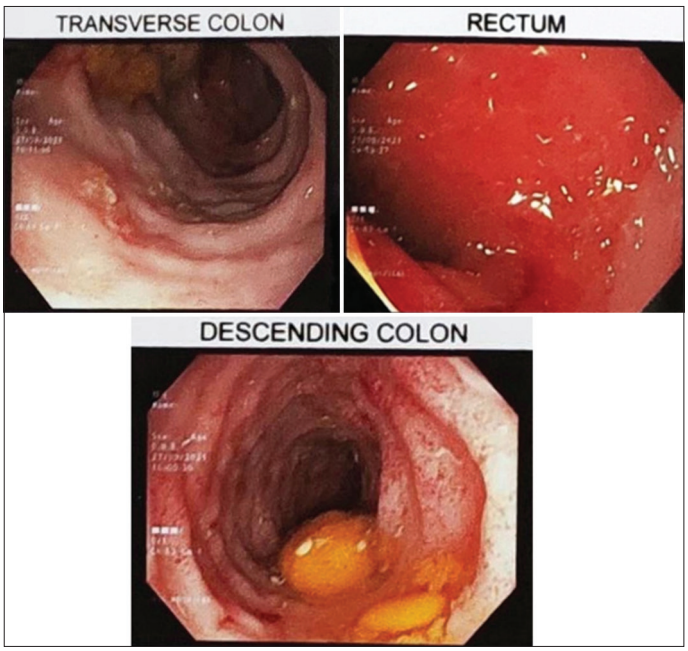

Figure 3: Colonoscopy findings in the transverse colon, rectum, and descending colon

mononuclear infiltrates and a few neutrophils. The fecal calprotectin level was elevated. Conclusively, a diagnosis of early-onset UC was made. As per the pediatric UC activity index (PUCAI) score [5] of 40/85, she suffered from moderate UC.

The child was started on oral Prednisolone (1 mg/kg/day) and Mesalamine (50 mg/kg/day), along with oral iron, folate, and other nutritional supplementation. The child improved symptomatically and she was discharged on the 10th day of admission. Steroid therapy was being tapered. However, the child was readmitted after 2 months with complaints of fever, blood-tinged loose stools, and abdominal pain. As stool microscopy revealed plenty of inflammatory cells, *Clostridium difficile* infection/amebic dysentery was suspected and she was treated with a course of intravenous antibiotics. Fever subsided but in view of persisting blood in stools, oral Azathioprine was started in addition to the ongoing Prednisolone and Mesalamine therapy. The child is on regular follow-up with us and is doing well on treatment with improved nutritional status.

DISCUSSION

IBD is caused by the dysregulated or inappropriate immune response to environmental factors in a genetically susceptible host [1]. UC is a subset of this chronic inflammatory disease of unknown etiology, characteristically localized to the large intestine, sparing the upper gastrointestinal tract. The inflammation is typically remitting and relapsing [3].

In children, IBD is divided based on the age of onset: Pediatric onset (<17 years), early onset (<10 years), very early onset (<6 years), infant/toddler onset (<2 years), and neonatal onset (<28 days) [1]. The prevalence of pediatric UC in Northern European countries and the United States varies from 100 to 200/100,000 population [3]. In India, the Colitis and Crohn's

Foundation registry found the incidence of UC in adults to be 6.02/100,000 [6]. However, there is a dearth of literature recognizing the disease burden in the pediatric population of the country. A high index of suspicion is required for the early diagnosis of UC in developing countries. In our case, there was an initial inclination toward local surgical causes, such as an anal polyp or solitary ulcers, or the anatomical anal defect. It was after investigating that IBD was later strongly considered. Yet, tuberculosis, a remarkable mimic had to be ruled out prior to initiation of therapy.

The common symptoms of UC include constipation, tenesmus, urgency, abdominal cramps, nocturnal bowel movements, and blood in stools [1]. It is difficult to suspect the disease in the acute or subacute (1–2 weeks) phase. The chronicity of symptoms (>2 weeks duration) will help clinch the diagnosis. Anorexia, weight loss, and malnutrition with growth failure may ultimately manifest if symptoms persist [1]. Fulminant colitis is defined as fever, severe anemia, hypoalbuminemia, leukocytosis, and >5 bloody stools per day for 5 days [1].

Several classification systems for UC are available, among which the PUCAI score [5] and montreal classification [7] for extent (E) and severity (S) of UC are well known. Extraintestinal manifestations, including skin and mucous membrane involvement, are well known in IBD and tend to be more common with UC than with Crohn's disease. This was evident in our child in the form of multiple hyperpigmented lesions over the trunk and extremities.

The effective management of pediatric IBD requires a multidisciplinary approach, involving a pediatrician, gastroenterologist, nutritionist, child psychiatrist, and surgeon [3]. At present, no permanent medical cure is available for UC [1] and the goals of treatment in children are directed at controlling symptoms of the disease with minimal adverse effects of the drugs and to achieve normal or near normal functioning of the patient. The treatment options include aminosalicylates, corticosteroids, immunomodulators, monoclonal antibodies, and surgery in complicated cases. Other supportive measures such as adequate hydration, treatment of anemia and other nutritional deficiencies, proper diet, and psychosocial support play a very crucial role. In the present case, a thorough physical examination revealed an anteriorly displaced anus which was strong suspicion of rectal bleeding in this child initially. Given the poor nutritional status, infections such as amoebiasis and tuberculosis, especially in India would be a strong possibility and hence, the diagnosis of IBD would be challenging.

CONCLUSION

Early-onset UC is an important emerging disease in the pediatric population posing a diagnostic challenge in a developing country like India where infectious diseases are prevalent and may mislead the diagnosis. Awareness of the fact that IBD can occur in children <10 years old will help in early recognition and prompt management of this condition.

REFERENCES

1. Kliegman R. Nelson Textbook of Pediatrics. 21st ed. Philadelphia: Elsevier; 2020. p. 1976-81.
2. Ruemmele FM, El Khoury MG, Talbotec C, Maurage C, Mougenot JF, Schmitz J, *et al*. Characteristics of inflammatory bowel disease with onset during the first year of life. J Pediatr Gastroenterol Nutr 2006;43:603-9.
3. Senbanjo IO, Oshikoya KA, Onyekwere CA, Abdulkareem FB, Njokanma OF. Ulcerative colitis in a Nigerian girl: A case report. BMC Res Notes 2012;5:564.
4. Banerjee R, Pal P, Nugent Z, Ganesh G, Adigopula B, Pendyala S, *et al*. IBD in India: Similar phenotype but different demographics than the west. J Clin Gastroenterol 2020;54:725-32.
5. Turner D, Otley AR, Mack D, Hyams J, De Bruijne J, Uusoue K, *et al*. Development, validation, and evaluation of a pediatric ulcerative colitis activity index: A prospective multicenter study. Gastroenterology 2007;133:423-32.
6. Sood A, Kaur K, Mahajan R, Midha V, Singh A, Sharma S, *et al*. Colitis and Crohn's foundation (India): A first nationwide inflammatory bowel disease registry. Intest Res 2021;19:206-16.
7. Satsangi J, Silverberg MS, Vermeire S, Colombel JF. The montreal classification of inflammatory bowel disease: Controversies, consensus, and implications. Gut 2006;55:749-53.

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