Case Report

Ulcerative Colitis in Infancy

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ABSTRACT

Ulcerative colitis (UC) is a chronic idiopathic inflammatory disorder of colon. Frequency of UC is gradually increasing over few years worldwide. Prevalence is 35 to 100/100 000 people in USA, 1% of them are infants. UC develops in a genetically predisposed individual with altered intestinal immune response. An eight-month-old girl presented with loose bloody stool, growth failure, and moderate pallor. The girl was diagnosed as a case of UC by colonoscopy and biopsy. Treatment was thereafter started with immunosuppressive drugs. After initial induction therapy with parenteral steroid and infliximab, the patient is now on remission with azathioprine and mesalamine. UC is rare in Bangladesh, especially in children, and it is rarer during infancy. Several conditions like infective colitis, allergic colitis, Meckel's diverticulitis, Crohn's disease, etc. may mimic the features of UC. So, if a child presents with recurrent bloody diarrhea, UC should be considered as differential diagnosis.

Key Word: Case report, infancy, ulcerative colitis

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Ulcerative colitis (UC) is a chronic, idiopathic, destructive disorder of colon. The annual incidence of UC is 10.4 to 12/100 000 people and prevalence is 35 to 100/100 000 people in USA.[1] Prevalence is lower in Asia.[2] UC seems to have a male preponderance. The incidence of UC peaks in the age group of 15 to 25 years and in 55 to 65 years, although it may occur at any age.[3] In patients with UC, 20% are younger than 20 years of age and 1% are infants.[4]

UC is a multifactorial, polygenic disease. It is now becoming clear that UC develops in a genetically predisposed individual with altered intestinal immune response.[5,6]

At the time of presentation, 44 to 49% of children with UC have rectosigmoid disease, 36 to 41% have left-sided disease, and 14 to 37% have pancolitis. In terms of severity of illness, 50 to 60% present with mild illness, 30% with moderate disease, and 19% with severe disease.[7] The cardinal symptoms of UC are diarrhea, rectal bleeding, abdominal pain, and weight loss. Extraintestinal manifestations develop in about 25 to 35% of patients. Extraintestinal manifestations may be the presenting features and may precede the gastrointestinal manifestation of UC.[8] Peripheral or axial arthritis is the most common extraintestinal manifestation occurring in about 7 to 25% of patients. Other extraintestinal manifestations include pyoderma gangrenosum, ocular complications, primary sclerosing cholangitis, nephrolithiasis, and pancreatitis.[9-11]

There is no available data regarding the incidence and prevalence of UC in Bangladesh. It is rare in children of Bangladesh. Though UC in infancy is rare and many conditions mimic UC, everybody should keep the suspicion of UC in mind if an infant presents with repeated diarrhea and bloody stool. If early pick up of disease is possible and treatment can be instituted earlier, outcome will be good and complications can be prevented.

CASE REPORT

A 14-month-old girl presented in Paediatric Gastroenterology and Nutrition Department of Bangabandhu Sheikh Mujib Medical University with complaints of loose motion with bloody stool since 8 months of age. Patient passed 4 to 8 stools per day. Blood was mixed with stool and mucus. Defecation was associated with tenesmus. Child was not growing well for the same duration. The patient had no

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history of fever, contact with tubercular patient, convulsion, respiratory distress or cough, oral or perianal ulcer, and major illness other than this. She was treated for diarrhea from the very beginning of illness with various antibiotics and drugs, but her condition was not improved with those treatments.

On physical examinations, the child was found ill looking, moderately pale, mildly dehydrated, moderately wasted, and stunted. She had nontender hepatomegaly. Otherwise, the child was normal.

The child was investigated and worked up for chronic diarrhea with blood in stool. Complete blood count showed Hb% - 8 gm/dl, with microcytic hypochromic anemia, total count of WBC was 8 500/cumm, and platelet count was 237 000/cumm. Gastrointestinal infections were ruled out by culture and sensitivity. Liver function tests and renal function tests were normal. Erythrocyte sedimentation rate was 53 mm/1st hour and C- reactive protein was 24 mg/l. Meckel's scanning was negative. IgM for cytomegalovirus and Herpes simplex I and II was negative. Serum immunoglobulin level was normal. Barium study of gastrointestinal tract was normal. At the age of 14 months, the patient underwent colonoscopy and biopsy. Colonoscopy showed diffuse ulcers and pseudopolyps formation in colon suggestive of UC [Figure 1]. Biopsy showed diffuse inflammation of mucosa and submucosa, crypt abscess, and depletion of goblet cells. Biopsy was also taken from stomach, duodenum, and rectum, but histologically was unremarkable. After diagnosis of UC at 14 months of age, the child was managed with dietary modification along with various antibiotics like metronidazole and ciprofloxacin, mesalamine, immunosuppressive therapy with methyl prednisolone, prednisolone, and azathioprine. Metronidazole at a dose of 30 mg/kg/day and ciprofloxacin at a dose of 20 mg/kg/day were given orally for 2 weeks. Mesalamine was added at a dose of 50 mg/kg/day orally. Four weeks after starting treatment, as the patient did not respond, methyl prednisolone intravenously followed by oral prednisolone were given at a dose of 2 mg/kg/day. Azathioprine was added at a dose of 2 mg/kg/day orally because the child was not responding to above mentioned drugs. Treatment was discontinued due to noncompliance of the parents. Then, at 21 months of age, treatment was again started with mesalamine, methyl prednisolone, prednisolone, and aziathioprine at the same manner, same dose, and same route. Due to failure to attain remission, cyclosporine was given orally at a dose of 4 mg/kg/day. Finally, infliximab was added at a dose of 10 mg/kg/dose parenterally and patient gradually improved with this treatment. Patient underwent remission 14 months after starting treatment. She is on remission now and it was sustained for last 9 months. Now, she is on mesalamine and azathioprine therapy for the last 3 months.

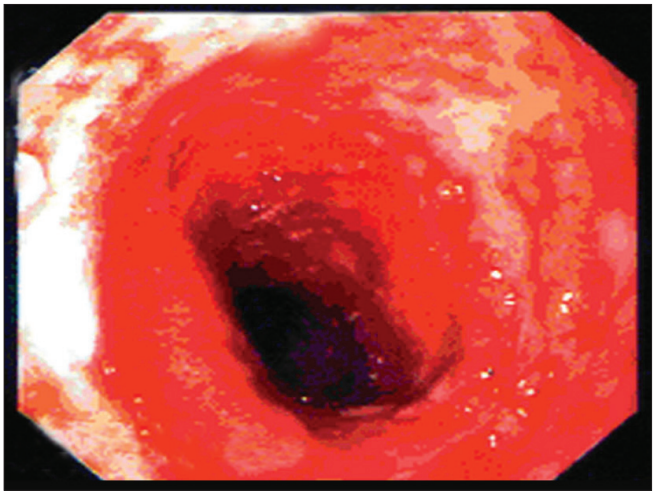

Figure 1: Image shows colonoscopic findings of ulcerative colitis

She was followed-up every two months. At follow-up, she was found good looking, gradually decreasing blood in stool, appetite and general condition improving, erythrocyte sedimentation rate and c-reactive protein gradually becoming normal. For the last 9 months, there is no report of blood in stool and stool is being passed normally.

DISCUSSION

UC occurs commonly in the age of 15 to 25 years and 55 to 65 years of age.[3] But, our case presented at 8 months of age. This age of presentation is extremely unusual. This early presentation indicates its genetic predisposition. Several genetic risk factors have been identified in UC. Few genetic markers on chromosome 1p36 and 12q15 showed highly significant associations with UC. In several studies, it was found that people not exclusively breastfed are at increased risk of developing UC.[12-14] Our case is an irregularly breastfed baby.

UC commonly occurs in male and Crohn's disease in female.[3] Most of the patients of UC presents with diarrhea, bloody stool, abdominal pain, and growth failure.[15] Our patient also presented with almost similar type of complaints.

UC may even present with constipation. Constipation may be observed in those with proctitis. Bloody stool is present in 90% patients.[16] Growth failure is present in 20% patients with UC.[17] Blood in stool is due to sloughing of mucosa and submucosa of colon.[5] Growth failure in UC is due to decrease in intake, chronic diarrhea, increased catabolism, and production of certain growth inhibitory cytokines.[18] Growth failure often is the initial presenting symptom in children with early-onset disease and can pose an ongoing problem during childhood, especially for those with severe and poorly controlled disease. Impaired linear growth occurs in 6 to 10% of children.[3]

The patient in this case report was moderately anemic. Anemia may result from chronic blood loss, decreased intake, and elaboration of inflammatory cytokines that can interfere with erythropoiesis.[14]

Several conditions may mimic UC. These are Crohn's disease, infective colitis, allergic colitis, Meckel`s diverticulum, intussusceptions, and immunodeficiency disorder.[1] Crohn`s disease was excluded by biopsy. Rarely UC may turn to Crohn's disease, but such changes may occur in older people, not in infancy. GIT infection was excluded by stool culture. Allergic colitis was also excluded by dietary modification. Common food allergens like animal milk, egg yolk, and soya protein were sequentially withdrawn from infant diet. Amino acid-based formula (Neocate) was used, but the patient did not improve to these dietary modifications. There was also absence of eosinophilic infiltrate in colonic mucosa in biopsy. Meckel`s scanning was negative. Intussusception was excluded by digital examination and imaging studies. Immunodeficiency disorders were excluded. Serum immunoglobulin levels were normal. There were no features of immunodeficiency disorder clinically. Moreover, secondary immunodeficiency like human immunodeficiency virus infection is very rare in Bangladesh.

A long list of tests can be done for the diagnosis of UC, but Kim and Ferry identified UC solely by clinical history (gastrointestinal bleeding and abdominal pain) and routine laboratory tests, and diagnosis was confirmed by colonoscopy and biopsy. There is little benefit in adding serological testing for diagnostic purpose.[3]

Evidence-based treatment of UC presenting in infancy is limited. The goal of medical therapy in UC in children include the induction of remission with control of symptoms, the prevention of relapse, the avoidance of complications, and the provision of optimal quality of life.[19,20] Therapeutic strategy have evolved from anecdotal experience and published series and case reports. In infants, a small number will respond to an elemental diet. Those unresponsive to nutritional therapy are customarily treated with steroids. Mesalamine can also be used in UC during infancy. Despite initial response to elemental diet, prednisolone and mesalamine, many patients eventually require 6-mercaptopurine, tacrolimus, azathioprine, and infliximab. In refractory cases, colectomy is needed.[21,22]

Mild UC can be treated with mesalamine. Moderate UC is treated with prednisolone, mesalamine, azathioprine, 6-mercaptopurine, or methotrexate. Infliximab and cyclosporine are used in addition to abovementioned drugs in severe UC.[1] Our patient was a case of severe UC and was treated accordingly. With this treatment, remission usually attains by 4 months.[7] In our patient, remission was attained by 14 months of treatment. Remission was assessed with clinical improvement and lowering down of erythrocyte sedimentation rate and c- reactive protein. Only 5% patients attain long-term remission.[5] Recurrence by 1 year has been reported in as high as 75% of patients.[11] Beyond the first decade of disease, the risk of developement of colon cancer begins to increase rapidly. Surgical management of UC is needed in some patients and indications include perforation, toxic megacolon, failure to medical treatment, dysplastic changes in colonic epithelium, or malignancy.[21] Ultimately, more than 50% of patients require surgical treatment.[22]

CONCLUSION

UC in first year of life is reported for the first time in Bangladesh and is rare in this age group worldwide. It is well known that UC is a disease of older children and adults, but if a patient presents with recurrent bloody diarrhea even in infancy, UC should be considered as a differential diagnosis. If proper investigations, appropriate treatment with immunosuppressive therapy, and meticulous follow-up can be done, a normal healthy life is possible in these children.

REFERENCES

1. Jang ES, Lee DH, Kim J. Age as a clinical predictor of relapse after induction therapy in ulcerative colitis. Hepatogastro-enterology 2009;56:1304-9.
2. Diefenbach KA, Breuer CK. Pediatric inflammatory bowel disease. World J Gastroenterol 2006;12:3204-12.
3. Kim SC, Ferry GD. Inflammatory bowel disease in pediatric and adolescent patients: Clinical, therapeutic and psychosocial considerations. Gastroenterology 2004;126:1550-60.
4. Kappelman MD, Graud RJ. Does inflammatory bowel disease develop in infants? Inflamm Bowel Dis 2009;15:1438-47.
5. Podolsky DK. Inflammatory bowel disease. N Engl J Med 2002;347:417-28.
6. Hugot JP, Bellaiche M. IBD: The pediatric gastroenterologist's perspective. Pediatr Radiol 2007;37:1065-70.
7. Baurngari DC. The diagnosis and treatment of Crohn's disease and ulcerative colitis. Disch Arztebi Int 2009;106:123-33.
8. Baud C, Saguintah M, Veyrac C. Sonographic diagnosis of colitis in children. Eur Radiol 2004;14:2105-19.
9. Baumgart DC, Sandborn WJ. Inflammatory bowel disease: Clinical aspects and established and evolving therapies. Lancet 2007;369:1641-57.
10. Baumgart DC, Carding SR. Inflammatory bowel disease: Cause and immunobiology. Lancet 2007;369:1627-40.
11. Xavier RJ, Podolsky DK. Unravelling the pathogenesis of inflammatory bowel disease. Nature 2007;448:427-34.
12. Rucmmele FM, ELKhoury MG, Talbotte C. Characteristics of inflammatory bowel disease with onset during the 1st year of life. J Pediatr Gastroenterol Nutr 2006;43:603-9.
13. Mamula P, Telega GW, Markowitz JE. Inflammatory bowel disease in children 5 years of age and younger. Am J Gastroenterol 2002;97:2005-10.
14. Barclay AR, Russel RK, Wilson ML, Gilmour WH, Satsangi J, Wilson DC. Systemic review: The role of breastfeeding in the development of paediatric IBD. J Paediatr 2009;155:421-6.

15. Hoddinott P, Tapin D, Wright C. Breastfeeding. Br Med J 2008;336:881-7.
16. Baron S, Truck D, Leplat C, Merle V, Gower-Rousseau C, Marti R. Environmental risk factors in pediatric inflammatory bowel diseases: A population based case control study. Gut 2005;54:357-63.
17. Marx G, Siedman EG, Martin SR. Outcome of cron's disease diagnosed before 2 years of age. J Pediatr 2002;140:470-3.
18. Hildebrand H, Finkel Y, Grahrquist L. Changing pattern of pediatric inflammatory bowel disease in northern stockholm 1990-2001. Gut 2003;52:1432-4.
19. Heyman MB, Kirschrer BS, Gold BD. Children with early onset IBD: Analysis of a pediatric IBD consortium registry. J Pediatr 2005;146:35-40.
20. Hyams JS. Chronic ulcerative colitis. In: Kliegman RM, Behrman RE, Jenson HB, Stanton BF, editors. Nelson Textbook of Pediatrics. Philadelphia: Saunders; 2007. p. 1577-80.
21. Croft NM. Ulcerative colitis. In: Kleinman RE, Sanderson IR, Goulet O Sherman PM, Mieli-Vergan G, editors. Pediatric Gastrointestinal Disease. Pennysylvania: BC Decker Inc; 2008. p 519-43.
22. Yang SK, Loftus EV, Sandborn WJ. Epidemiology of inflammatory bowel disease in Asia. Inflamm Bowel Dis 2001;7:260-70.

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