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CASE REPORT

Infantile Inflammatory Bowel Disease in A 14 Month Old Bangladeshi Boy: An Unusual Presentation

Tanzila Farhana[1], Salahuddin Mahmud[2], Mashud Parvez[3], Madhabi Baidya[4], Ahmed Rashidul Hasan[5], Syed Shafi Ahmed[6]

Introduction

The term inflammatory bowel disease (IBD) is used to represent two particular disorders, Crohn's disease (CD) and Ulcerative colitis (UC). It has been pointed out that; the frequency of childhood onset inflammatory bowel disease (IBD) is increasing throughout the world. P-IBD (onset before 17 years of age) has been sub-classified based on the age of onset of the disease as neonatal onset (<28 days of life), infantile onset (<2 years of age), very early onset (<6 years of age), and early onset (<10 years of age).[1,2] The incidence of paediatric onset UC, which constitutes roughly 15% to 20% of all UC, ranges at 1to 4/ 1000000/ year in most North American and European regions.[3] A study of Canada showed the incidence of IBD increased from 9.4 per 100,000 children (95% confidence interval [CI], 8.2-10.8/ 100,000 children) in 1994 to 13.2 per 100,000 children (95% CI, 11.9-14.6/100,000 children) in 2009 ($P<$ .0001). The incidence increased by 7.4% per year among children younger than 6 years old and 6-9.9 years old, and by 2.2% per year among children ≥10 years old.[4] Nevertheless, in the countries of Asian subcontinent like Bangladesh, VEO-IBD patients have not been extensively studied, mostly because of the rarity of this patient population. So there has been remained a scarcity of research, in order to obtain a better sense of what they respond to and what they don't. The presentations of childhood-onset IBD are more aggressive, rapidly progressive and often needed early surgical interventions as compared to the characteristics of the adult cohort.[5-7] Infantile IBD should be considered as an important differential diagnosis of any child <2 years of age presents with persistent diarrhea, abdominal pain, hematochezia, failure to thrive, and/or poor feeding. Assessing the severity of the disease greatly relies on The Pediatric Ulcerative Colitis Activity Index (Table II) is an authentic score of clinical disease activity, not required endoscopy or laboratory markers and is accessible to calculate routinely. A clinically significant response is indicated by a decline in PUCAI of at least 20 scores. Moreover in drug trials, the PUCAI score is advantageous, including

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1. Resident Medical Officer, Department of Paediatric Gastroenterology, Hepatology & Nutrition, Bangladesh Shishu Hospital & Institute, Dhaka.
2. Associate Professor, Department of Paediatric Gastroenterology, Hepatology & Nutrition, Bangladesh Shishu Hospital & Institute, Dhaka, Bangladesh.
3. Associate Professor, Department of Pathology, Bangladesh Shishu Hospital & Institute, Dhaka, Bangladesh.
4. Assistant Professor, Department of Paediatric Gastroenterology, Hepatology & Nutrition, Bangladesh Shishu Hospital & Institute, Dhaka, Bangladesh.
5. Registrar in-charge, Department of Paediatric Gastroenterology, Hepatology & Nutrition, Bangladesh Shishu Hospital & Institute, Dhaka,
6. Professor and Head, Department of Pediatric Gastroenterology, Hepatology & Nutrition, Bangladesh Shishu Hospital & Institute, Dhaka, Bangladesh.

Correspondence to: Dr. Tanzila Farhana, Resident Medical Officer, Department of Pediatric Gastroenterology, Hepatology & Nutrition, Bangladesh Shishu Hospital & Institute, Dhaka, Bangladesh. Cell: +8801737257050, E-mail: tanzila.djmc@gmail.com

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high correlation with colonoscopy. Here the authors will report case of a 14 month-old Bangladeshi boy who presented with bloody diarrhea and was finally diagnosed as Infantile IBD and treated conservatively. He showed acceptable improvement and was followed up, up to a certain period of time.

Case Report

A 14 month old boy was referred to the Department of Pediatric Gastroenterology, Hepatology and Nutrition unit of Bangladesh Shishu Hospital & Institute, having frequent passage of blood mixed loose stool for last 7 days. The frequency was about 10-12 times per day and other associated conditions were, low grade intermittent fever, occasional abdominal pain and irritability. In last 2 months preceding hospitalization, he had experienced off and on passage of loose stool mixed with mucus but no blood. His medical history indicated the ante natal, natal and post-natal period was uneventful. He was on exclusive breast feeding up to 6 months of his age later complimentary feeding was started with family foods but no allergic ones (like cow's milk, nuts etc.) was introduced. For these reason he was treated several times by different physicians without any long lasting improvement.

On the day of admission, the boy passed about 10 bowel movements a day, which was bloody and >50% of the stool volume being blood and it persisted for next 7 days. Many of the purging occurred at night. It was associated with abdominal pain as because the boy was crying during purging. On general examination, he looked irritable, mildly pale, anicteric, febrile, temperature was 100°F, heart rate was 124/min, blood pressure was 90/60 mmHg, respiratory rate was 18 breathes/min. There was no lymphadenopathy or skin lesion present. His height was 78cm (falls on 50th centile; ref. value CDC) and weight was 10 kg (falls on 10th centile; ref. value CDC).

On systemic examination, abdomen was soft, mildly distended, no rigidity, ascites, gurding or organomegaly was present. No abnormality was seen on the perianal region and other systemic examination revealed normal finding. With the aim of precluding the differential diagnosis, microbiological testing was done. Routine microscopic examination of stool showed us there was presence of plenty of pus cells along with numerous RBC and Stool for OBT found positive. Stool and Blood cultures were found no growth of any organism. Allergic testing was also performed and found normal. The results of laboratory tests are shown on *Table 1.* Immunoglobulin levels with flow cytometry were normal in range and HIV testing was also done which found negative. The abdominal Ultrasonography and Chest X-ray was normal. We also did RT PCR for COVID-19 (found negative) before performing the upper GI endoscopy and Colonoscopy.

Table I
Laboratory results

Parameters	Initial values	F/up values	Normal range/unit
Hemoglobin	9.2	9.8	11.5-14.5gm/L
WBC count	18,700	8,500	6,000-17,500 /mm^3
Platelet count	506,000	365,000	1, 50,000-4, 50,000 / mm^3
N%	45	38	28
L%	44	58	61
M%	06	03	3
E%	05	01	5
ESR	68	35	
Na+	145.0	136.5	135-156 mmol/L
K+	3.6	4.5	3.5-5.4 mmol/L
Cl^-	105.0	104.0	96-108 mmol/L
S. creatinine	64.0	-	40-110 umol/L
S. Ca++	2.1	-	2.02-2.6 mmol/L
S. Albumin	32.9	38.7	35-60 gm/L
D-Dimer	2.5	1.2	<0.5 mg/L
CRP	116.0	12.6	<5 mg/L
ANA	Negative	-	
PT	10.7	-	12 sec
F. calprotectin	890.0	95.0	<80 µg/gm
IgE	10.0	-	<29 IU/ml

Colonoscopy report showed, the mucosa of the rectum, sigmoid colon, descending colon and up to mid transverse colon was inflamed and friable. Vascular pattern was lost in the affected segment. The mucosa, vascular pattern and lumen of the rest of the colon appeared normal including the anal canal (*Figure 1a, 1b, 1c*). Biopsy specimen was taken from inflamed area of the colon and histopathological report revealed, colonic tissue presenting ulcerations, edema and acute inflammatory exudates. The lamina propria was edematous with cryptitis. Crypt abscess and diffuse mixed inflammatory cellular infiltrates predominant cells were lymphocytes, plasma cells, polymorphs and

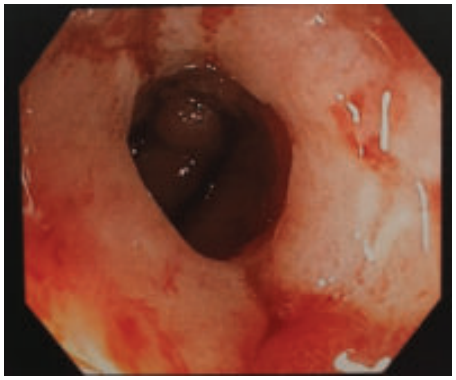

Fig.-1a *Inflamed mucosa*

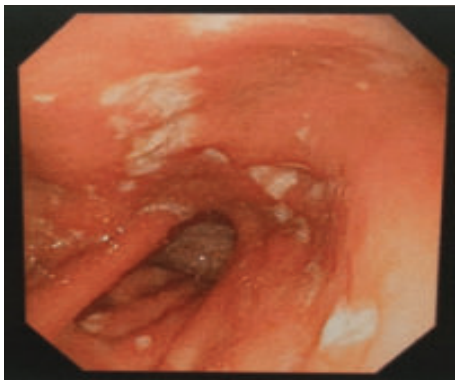

Fig.-1b *Scattered ulceration*

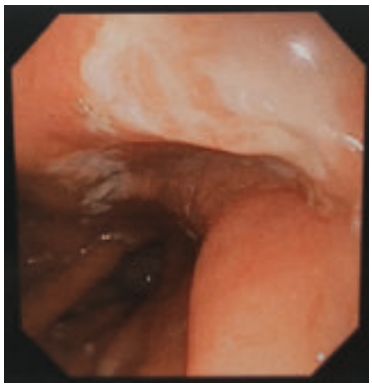

Fig.-1c *Large ulceration*

Fig.-1(a,b,c) *Colonoscopic view of infantile IBD*

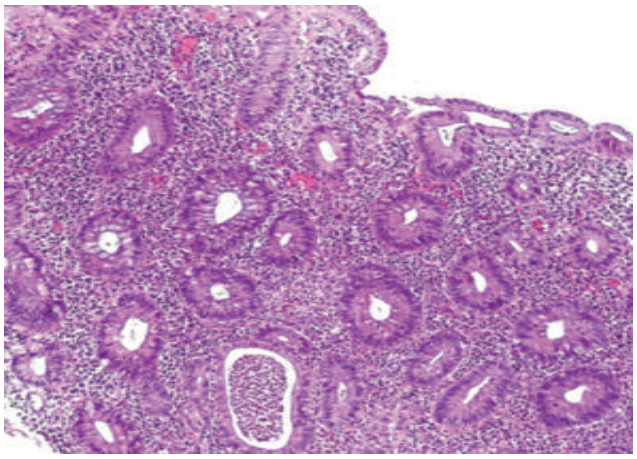

Fig.-2: *Colonic tissue, H & E stain low-power image reveal crypts abscess and cryptitis*

eosinophils (Figure-2). No architectural distortion or basal plasmacytosis was seen. Endoscopy of upper GIT revealed normal finding. Disease activity was assessed as 70 Score on the PUCAI scale (Paediatric Ulcerative Colitis Activity Index Table II). Ulcerative colitis was the final diagnosis. Treatment was planned with IV Methylprednisolone 1.5mg/kg/day for 5 days; along with broad spectrum antibiotic inj. Meropenem. We also restricted feeding initially and gradually increased the amount with continued breast milk and rice suji accordingly. From 1st week of treatment we observed gentle clinical improvements; the numbers of bowel movement were ceased, stool no longer contained blood, no night time purging, baby became playful, and fever subsided.

Table II
PUCAI score

Paediatric Ulcerative Colitis Activity Index (PUCAI)			
1. Abdominal pain		***4. No. of stools***	
No pain	0	0-2	0
Pain can be ignored	5	3-5	5
Pain cannot be ignored	10	6-8	10
2. Rectal bleeding	>8	15	
None	0	***5. Nocturnal stools***	
Small amount only in<50% stools	10	No	0
Small amount with most stools	20	Yes	10
Large amount >50%of the stool content	30	***6. Activity level***	
3. Stool consistency		No limitation	0
Formed	0	Occasional limitation	5
Partially formed	5	Severely restricted activity	10
Completely unformed	10		

PUCAI Scoring with interpretation			
<10	Remission	**10-34**	Mild disease activity
35-64	Moderate disease activity	**>65**	Severe disease activity

At this point of treatment we switched to oral prednisolone 1mg/kg/day and Mesalazine was added (40mg/kg/day). One week after discharge on his 1st follow-up visit PUCAI score was 5. General condition was good, he regained appetite. He was passing 2 bowel movements per day, which was of normal consistency with no blood or mucus. CBC, CRP, S. albumin levels showed significant improvements.

It was recommended to reduce the oral dose of Prednisolone gradually, to continue Mesalazine, to expand the range of the diet slowly added some adjunctive medications like Zinc, Folic acid, Cholecalciferol and he was scheduled for regular follow-up visit.

Discussion

It is distressing that the incidence of Pediatric IBD is inflating worldwide, having a multifactorial etiopathogenesis. Our patient presented at his 14 months of age, Sara Ebrahimi reported a neonate who presented at 7 days of life and Chie Iida reported of an infant of 3 months of age.[8, 9]

In our case symptoms were found persistent diarrhea, abdominal pain, hematochezia, irritability, and poor feeding, similarly Xin Wang and Yuan Xiao found, patients in the 0–2 y group commonly manifested systemic symptoms such as fever, weight loss, diarrhoea and limitation of activity, colonic lesions, strictures and perianal disease. [10-13] The prevalence of extra-intestinal manifestations in IBD reportedly ranges from 6% to 47%. But in our case there was no such type of involvements like perianal disease. Our patient's PUCAI score was above 65 which indicated severe form. [14-17] The laboratory findings included mild anemia, leukocytosis and thrombocytosis in CBC, a raised ESR & CRP with a normal IgE level and primary immunodeficiency panel. It was almost similar with the findings of Ahamed N [18], so that other differentials like milk protein allergy, primary immunodeficiency syndrome were excluded. At this age it has a strong connection with IL-10 receptor gene, unfortunately we didn't have access for gene analysis which can be a limitation of our study. Colonoscopic and Histopathological evidences confirmed the diagnosis of UC. We initially administered IV Methylprednisolone 1.5mg/kg/day for 5 days; along with broad spectrum antibiotic inj. Meropenem. When purging rate was declining as well as the clinical features were improving we switched over to oral prednisolone 1m/kg/day later it was tapered within 2.5 months and Mesalazine was added (40mg/kg/day), as there are many side effects such as growth disturbances, osteoporosis, immunosuppression are documented in the long term use of steroids. Our patient responded gradually with anti-inflammatory agents and antibiotics and we discharged him with the plan of regular follow-up.

Conclusion

Chronic bloody diarrhea at any age should be evaluated properly for IBD. Colonoscopy and histopathology should be done routinely to make a diagnosis. PUCAI scoring can be used to measure the severity of the disease. As the infantile IBD has a poor prognostic factor so combined therapy should be started as early as possible to prevent surgical intervention.

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