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Case Report

Challenges in Diagnosis and Treatment of a Child with Inflammatory Bowel Disease in Resource-Limited Setting - A Case Report

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Abstract

Inflammatory Bowel Disease (IBD) is not commonly encountered in children in sub-Saharan Africa. It is reported mostly in industrialized countries. This is a report of a 12-year old Nigerian boy with chronic diarrhoea and undernutrition. Common causes of diarrhoea and undernutrition were excluded. There was however difficulty in confirming the suspected IBD due to lack of diagnostic investigation facilities in the region. Open surgical biopsy of the intestine was done and the histological features suggested Crohn's disease. The child responded to oral steroid treatment. It is recommended that IBD be considered in children with chronic diarrhoeal and/or undernutrition especially when common causes are excluded. .

Keywords: *Crohn's Disease; Inflammatory Bowel Disease; Chronic Diarrhoea; Children; Nigeria*

Background

Inflammatory Bowel Disease (IBD) is a chronic relapsing gastrointestinal disorder. The main types of IBD are Crohn's disease (CD), ulcerative colitis (UC) and IBD unclassified (IBD-U) [1,2]. Crohn's disease is characterized by inflammation of the mucosa of the digestive tract, but may involve the deeper layers. The lesion may be located anywhere between the mouth and anal orifice [2]. It is due to immune dysregulated response to the intestinal microbiome and environmental factors in a susceptible host [3,4]. Children with CD have chronic diarrhoea, abdominal pains and commonly become undernourished [2].

The prevalence of CD in children is rising in both developed and developing countries [5-7]. The prevalence of IBD in Nigeria is not known. However, few cases were reported [8-11]. The apparent rarity of IBD in Nigeria may be due to challenges in confirming the diagnosis because of unavailability of diagnostic facilities. We present a case of typical challenges in diagnosing IBD in children in resource-limited setting.

Case Presentation

The patient was a 12-year-old male who presented at the Paediatrics Department of a University Teaching Hospital, in north eastern Nigeria with diarrhoea and growth failure of three years duration. Each episode of diarrhoea lasted for days to weeks, with diarrhoea-free interval of three to five days. The diarrhoea was watery, non-bloody, not frothy, not oily and of large volume. No report of seeing undigested food material in the stool. He was having 4 to 10 episodes per day, and it was not modified by type of food he ate. There was

associated occasional central abdominal pain. No abdominal distension. His appetite for food was preserved and he consumed family diet which was fairly adequate.

He had not been growing adequately since the onset illness, and had been less active but not bed ridden. There was no associated fever, cough or body swelling. No contact with persons with chronic cough. He had BCG vaccine in infancy. His parents were native of north eastern Nigeria. He was the first of family's three children. No one else in the family with similar complaints.

He had been to many healthcare facilities since onset of the illness, and had taken many medications which included oral rehydration salt (ORS) solution, oral zinc tablets, anti-helminthic medications, and several antimicrobials without resolution of the symptoms.

On examination, he was chronically ill-looking, pale, and had a normal axillary temperature of 36.8°C. There was mild dehydration. He had no oedema, no jaundice, no cyanosis, and no significant lymph node enlargement. His weight progressively declined from 31 kg to 26.5 kg. He was severely wasted (BMI 12.87 kg/m^2), and stunted with height of 143.5 cm (15th percentile of WHO child growth standard).

He had dry lips with linear ulcers on them and angular stomatitis. His abdomen was flat, soft and no area of tenderness. His liver was palpable 4cm below the right costal margin, soft, smooth and not tender. No other masses were palpated. Ano-rectal examination revealed clean anal area and good anal sphincter tone. The rectum was empty and had smooth nontender surface. No blood was seen on the gloved finger.

Investigations

Stool microscopy showed pus cells, and ova of *Ascaris lumbricoides*. No bacterial or protozoan cells were isolated. Screening for human immunodeficiency virus (HIV) infection and mycobacterium tuberculosis (GenXpert, Mantoux test, chest x-ray, abdominal ultrasound scan) were negative. His serum proteins were low (total 5.3 g/dL; albumin 2.4 g/dL). Liver aminotransferases and serum bilirubin levels were normal. Serology for HBsAg was non-reactive. His electrolytes panel showed hypokalaemia. Assay for anti-*Saccharomyces cerevisiae* antibody (ASCA), peri-nuclear antineutrophil cytoplasmic antibody (pANCA) and faecal calprotectin were requested but could not be done due to non-availability of the tests.

Laboratory investigation	Test result	Remarks
Stool microscopy	Ova of *Ascaris lumbricoides*	Abnormal
ELISA for HIV	Negative	Negative
GeneXpert	No MTB	No mycobacteria detected
Mantoux test	Non-reactive	Non-reactive
Total serum proteins (g/L)	53	Low
Serum albumin (g/L)	24	Low
AST (IU/L)	10	Normal
ALT (IU/L)	12	Normal
Total bilirubin (µmol/L)	3.5	Normal
Conjugated bilirubin (µmol/L)	0.3	Normal
Serum sodium (mmol/L)	120	Low
Serum potassium (mmol/L)	1.8	Low
Serum chloride (mmol/L)	75	Low
Serum urea (mmol/L)	2.7	Normal
Serum creatinine (µmol/L)	70	Normal

Table 1: *Laboratory investigation results.*

Due to non-availability of paediatric GI endoscopy facilities, surgical open intestinal biopsy was done. Histologic finding of the biopsy specimen showed isolated focus of villous blunting in the small intestine. There were also intra-epithelial lymphocytosis and presence of lymphocytic infiltrates in the lamina propria forming lymphoid aggregates. There was no focal crypt hyperplasia.

Based on the above clinical manifestations, small intestinal histologic features and laboratory findings, a diagnosis of chronic diarrhea secondary to Crohn disease was made. Other conditions such as Celiac disease and malabsorption syndromes were also considered.

Treatment

The child was initially resuscitated: oral rehydration salt (ORS) solution, oral zinc 20 mg daily, potassium tablets, and oral micronutrients preparation were administered. *Ascaris lumbricoides* infection was treated with oral albendazole 400 mg single oral dose. Repeat stool microscopy after this treatment was normal. He was also commenced on adequate diet. Despite this treatment the frequency and volume of diarrheal stool remained unchanged. He continued to lose weight and became progressively weak. Subsequently, a course of oral prednisolone 1 ml/kg/day for two weeks was given. Following this treatment, the frequency of diarrhoea was reduced to 1 to 2 times per day, and the volume of stool also reduced. He remained in remission until he was lost to follow up due to relocation of the parents to another state.

Discussion

Inflammatory bowel disease (IBD) is commonly seen in industrialized countries where its prevalence was up to 505 per 100 000 for ulcerative colitis, and 322 per 100 000 for Crohn's disease [5], IBD occurs less frequent in children. The highest annual incidences of paediatric-onset IBD were 23/100000 person-years in Europe, 15.2/100000 in North America, and 11.4/100000 in Asia/the Middle East and Oceania [12]. The actual incidence of IBD in Africa is not known [9]. IBD is believed to be uncommon in Africa, particularly sub-Saharan Africa where the rate of poverty is very high. In South Africa the incidence of Crohn disease was 0.3 - 2.6 cases per 100,000 persons [9]. Few Cases of IBD were reported from the southern part of Nigeria [8-11]. However, the disease may have been missed in many children in Africa due to partly lack of adequate clinical suspicion on the part of clinicians, and partly due to lack of appropriate diagnostic investigation facilities. Moreover, diarrhoea and undernutrition are among the common clinical manifestations of IBD, both of which are very common among children in Africa. This is illustrated in this patient who had been having diarrhoea for three years before he was eventually seen at the tertiary referral health centre. This delay presentation at the referral hospital might be due to attempts to diagnosed and/or treat more common illnesses in this region such as abdominal tuberculosis, intestinal protozoan infestations and bacterial infections that present with chronic diarrhoea. Even at the teaching hospital where IBD was suspected, these common illnesses were considered first and ruled out. Even though there are clear guidelines on diagnosis of IBD in children and adolescents [13,14], strict implementation of the guidelines is a huge challenge in developing countries such as Nigeria at this time. It is therefore important to strongly consider IBD in a child with chronic diarrhoea and failure to thrive despite adequate food intake when common causes are ruled out.

It is noteworthy that open full thickness intestinal biopsy was done to confirm the diagnosis because less invasive paediatric upper gastrointestinal endoscopy and colonoscopy facilities were not available. Similar findings were reported in the past in sub-Saharan Africa [9]. This is a huge limitation for the diagnosis of IBD and other paediatric gastroenterological diseases that need endoscopy and biopsy. There is an urgent need for investment in endoscopy machines and training of Paediatrician on endoscopy. In some cases of IBD reported from southern Nigeria [8-11], GI endoscopy and biopsy were done because they were available close by, and the patient could afford them. Other laboratory investigations that assist in making the diagnosis such as anti-Saccharomyces cerevisiae antibody (ASCA), peri-nuclear antineutrophil cytoplasmic antibody (pANCA) and faecal calprotectin were not available to our patient. However, in earlier report from Nigeria [11], some of these laboratory investigations were available, and their patients were able to afford them. Imaging techniques such as computed tomography (CT) and magnetic resonance enterography (MRE) were too expensive for patients in resource-limited

countries. Other imaging techniques that could be useful in such patients include wireless capsule endoscopy, small intestinal contrast ultrasound scan (SICUS), and balloon-assisted enteroscopy [1,2]. In some low income countries, including some sub-Saharan African countries the diagnosis of IBD is based on clinical findings only.[15] In contrast, children with suspected IBD in developed countries have access to these diagnostic investigations and imaging techniques.

Good response to oral steroid (prednisolone) is further supporting the diagnosis of Crohn disease in this patient. But prolonged or repeated exposure to steroids carries a high risk of developing complications of these drugs. Other drugs that can be used to treat IBD such as 5-Aminosalicylic acid derivatives (sulfasalazine), immunomodulators (cyclosporine, 6-mercaptopurine, methotrexate), and biologics (such as infliximab) are still not readily available or are not affordable by the patients in our setting.

Therefore, in a situation where challenges in making diagnosis of IBD are daunting, a child with chronic diarrhoea may be considered to have IBD when other common causes are excluded. In such situations, an empiric treatment for IBD with available medications may be commenced, and the response and drugs side effects closely monitored.

Conclusion

IBD may occur in children in northern Nigeria but confirming the diagnosis and availability of recommended medications are huge challenges.

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Conflict of Interest

None.

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