Atypical Presentation of Celiac Disease: Recurrent Acute Small Bowel Obstruction

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ABSTRACT

CONTEXT: Intussusception is the most common cause of small bowel obstruction in children under 4 years of age. Intussusception is not a widely recognized complication of celiac disease.

CASE REPORT: We present a clinical case of a 23-month-old boy with a 1-month history of watery diarrhea complicated by 2 episodes of intestinal obstruction, both had required surgery. He presented with acute and severe abdominal distention with bilious vomiting, and an appearance of intussusception on abdominal ultrasound. Upon further investigation, the diarrhea was found to be malabsorptive. The diagnosis of celiac disease was confirmed by the presence of specific serum autoantibodies (IgA Tissue transglutaminase and endomysium Antibodies >200 UI/ml with normal serum IgA level). He started a gluten-free diet and his symptoms were almost completely resolved.

CONCLUSION: Recurrent intussusception may be associated with celiac disease, so celiac serology is recommended in children with recurrent intussusceptions. However, intestinal tuberculosis and lymphoma associated with enteropathy should be considered in the differential diagnosis. Intussusception in celiac disease is usually transient and should be managed expectantly rather than early surgical reduction.

KEYWORDS: Recurrent small bowel obstruction, intussusception, celiac disease

Introduction

Intussusception is the most common cause of small bowel obstruction in children under four years of age. Most cases of intestinal intussusceptions, in a child with normal nutritional status, are idiopathic. However, if the clinical presentation is unusual, this may prompt further investigation.1,2

Our case report describes the recurrent intussusception with a history of chronic diarrhea, which is an unusual presentation in children with celiac disease.

Recurrent intussusception as a presenting symptom of pediatric celiac disease has been very rarely reported.3

Case

Twenty-three-month-old infant had presented with a history of liquid diarrhea over the previous 25 days, complicated by peripheral edema 2 days after onset, all evolving in a context of apyrexia and deterioration of general condition.

The clinical examination found, a lethargic infant, pale conjunctiva with edema on bilateral legs. He was afebrile with normal haemodynamic status. The adipose tissue was diminished in all levels. His weight was 8 kg (67 percentile) and his height was 81 cm (93 percentile). No masses, hepatomegaly, or splenomegaly were present.

Results of laboratory studies were as follows: Anemia at 9 g/dl, low serum ferritin at 10 ng/ml (30-400 ng/ml), hypoalbuminemia at 21 g/l (35-52 g/l), hypoproteinemia at 42 g/l (64-83 ng/ml), and low plasma cholesterol at 0.94 g/l (1.54-2.01 g/l). The other investigations showed a negative septic screening.

Three days later, bilious vomiting with abdominal pain and distension developed. Abdominal x-rays without preparation showed multiple abnormal hydroaeric levels (Figure 1). Abdominal ultrasound showed an excessive meteorism with dilated bowel loops and there were 2 target signs; 1 in the right hypochondrium measuring 2.33 × 2.89 cm (Figure 2) and the second in the right iliac region measuring 1.75 × 1.81 cm. This finding indicated that there might be double simultaneous intussusceptions (Figure 3).

Abdominal computed tomography (CT) was performed to rule out an organic origin of intussusception. The CT scan objectified entero-enteric intussusceptions extended from the sub-hepatic region to the right iliac region, with a slight enhancement, with coelio-mesenteric and lumbo-aortic lymph nodes measuring 3.9 × 2.5 cm, enhanced after contrast agent administration.

The intraoperative findings showed an ileo-ileo intussusception, the surrounding bowel appears normal without signs of inflammation or ischemia, with multiple mesenteric lymphadenopathy. The intussusception was successfully cured by manual reduction and the mesenteric lymph node biopsy revealed only reactive lymph-node hyperplasia eliminating a secondary cause particularly intestinal tuberculosis and T cell lymphoma.

After an initial resolution of symptoms, the child had presented on the third postoperative day another occlusive episode...
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with recurrence of bilious vomiting. The abdominal ultrasound confirmed the diagnosis of recurrent intussusception on the same lead point of the first. The child was operated 1 more time.

In the presence of anemia with low serum ferritin, hypoalbuminemia, hypoproteinemia and low plasma cholesterol we concluded that the diarrhea was malabsorptive. Diagnosis of celiac disease was confirmed by the presence of specific serum autoantibodies (IgA Tissue transglutaminase and endomysium Antibodies >200 UI/ml with normal serum IgA level). He started a gluten–free diet and his symptoms were almost completely resolved.

Discussion
Intussusception is a common gastrointestinal emergency in pediatric patients and it is normally idiopathic, usually following a viral infection. Its classical symptoms include acute abdominal pain, red currant jelly stools and abdominal mass in a child with normal nutritional status.1,4

The first-line investigation for diagnosis of intussusception in children is abdominal ultrasound, given its high sensitivity (98%-100%) and specificity (88%-100%).5

If the clinical presentation is unusual, the child has chronic diarrhea, recurrent intussusception or deterioration of general condition, further investigation should be conducted after treating the intussusceptions.1 In a child with chronic diarrhea and recurrent intussusception intestinal tuberculosis and T cell lymphoma associated with enteropathy should always be excluded, as was done in our patient.

In this case, celiac disease was the pathological condition behind the findings, with clinical presentation of recurrent intussusceptions. The correlation between celiac disease and recurrent intussusception is obvious; the symptoms disappeared after the patient was started on a gluten–free diet. In patients with recurrent intussusception celiac disease should be ruled out.

The cause is unknown but it has therefore been suggested that undiagnosed celiac disease is characterized by small bowel inflammation and will sometimes cause small bowel wall edema, intestinal lymph node swelling and dysmotility but also ulcers and strictures. This, in turn, could be the lead point for intussusception, which can develop singly or multiply.6

Another particularity is that intussusception in celiac disease is usually transient. Non-operative reduction has a high success rate and low complication rate. However, operative intervention may still be needed when there are complications, a palpable abdominal mass or when non-operative intervention is unsuccessful.7

Conclusion
Recurrent intussusception may be associated with celiac disease, so celiac serology is recommended in children with recurrent intussusception. However, intestinal tuberculosis and T cell lymphoma associated with enteropathy should be considered in the differential diagnosis. Intussusception in celiac disease is usually transient and should be managed expectantly rather than early surgical reduction.

Author Contributions
REQ wrote the manuscript, AL I wrote the manuscript with his support, HN I wrote the manuscript with her support, IA
supervised the findings of this work, AB supervised the findings of this work.

**Informed Consent**

Informed consent has been obtained from the patient’s parents for publication of the case report.

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