

CASE REPORT

Multiple small bowel intussusceptions as a feature of celiac disease

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Abstract

Intussusception is commonly associated with celiac disease in adults. However, it is rarely reported in children in radiology journals. Here, we report a case of a 3-year-old girl with celiac disease presented with complaints of intermittent abdominal pain, distension, and vomiting for 6 months. The patient was underweight (8.6 kg). Her X-ray of the abdomen standing revealed abnormal air-fluid levels and ultrasound of the abdomen revealed single small bowel intussusception. Contrast-enhanced CT abdomen done before planning surgery and it revealed five small bowel intussusceptions with few dilated small bowel loops. Her IgA antibodies to tissue transglutaminase were done to look for the cause of failure to thrive and its titer raised significantly. Gluten-free diet was started for her and symptoms were resolved within 7 days without surgical management.

Key words: Celiac disease; failure to thrive; multiple intussusception

Introduction

Intussusception is the most common cause of small bowel obstruction in children between the ages of 6 months and 4 years. Most (over 90%) have no lead point and are due to lymphoid hypertrophy, usually following a viral infection. Other lead points are Meckel's diverticula, adhesions, adenitis, trauma, celiac disease, duplications, lipoma, and inflammatory lesions; small bowel malignancy (either primary or metastatic) may account for less than one-third of adult intussusceptions. The image-guided reduction can be performed using a pneumatic technique or by contrast enema, under fluoroscopy or ultrasound guidance.

Case History

A 3-year-old girl was brought to the department of pediatrics with a complaint of intermittent abdominal pain

for 6 months, vomiting, and increased frequency of stool for 1 month and fever for 7 days. There was a history of similar complaints in the past.

On examination, the patient was cachexic and underweight (8.6 kg). Tongue, nail, and conjunctiva were pale with edema on bilateral legs. In the abdomen examination, there was abdominal distension; however, no evidence of hepatosplenomegaly and central nervous system (CNS), cardiovascular system (CVS), and respiratory system (RS) examinations were normal.

Total leucocyte count was 6200/cmm (within normal limits) and hemoglobin was reduced (10.7 g%). Serum sodium and potassium were reduced (130 mmol/dL and 1.7 mmol/dL, respectively). Renal and hepatic function tests were within normal limits.

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X-ray abdomen erect was showing multiple abnormal air-fluid levels in the abdomen [Figure 1]. Ultrasound findings were showing excessive gaseous abdomen with few dilated bowel loops and single intussusception in the left upper abdomen; however, no lead point of intussusception was identified [Figure 2]. Due to severe abdominal pain, a CT of the abdomen was done to look for bowel ischemia secondary to bowel obstruction and to look for a lead point of intussusception if any. CT was showing a total of five small bowel intussusceptions [Figure 3].

Her IgA antibodies to tissue transglutaminase were done to look for the cause of failure to thrive and its titer raised significantly.

Electrolyte imbalance was corrected, a gluten-free diet with nutritional supplements for anemia and malnutrition was started and symptoms were resolved within 7 days without surgical management. A follow-up ultrasound of the patient was done and did not reveal any abnormality.

Discussion

Intussusception as a presenting symptom of pediatric celiac disease has been very rarely reported.^[1] Although intussusception is the most common cause of intestinal obstruction in children, celiac disease is a frequent small bowel disease they rarely had been reported in association with each other.^[2]

In most children (90%) with intussusception, the lead point could not be found. But it is due to lymphoid hypertrophy, usually following a viral infection. Other lead points in children can be Meckel's diverticula, adhesions, trauma, celiac disease, enteric duplications, lipoma, and inflammatory polypoidal lesions.^[3]

In the past decades when barium studies were used more commonly in the diagnosis of malabsorption, transient intussusception was seen in 20% of patients with proven celiac disease.^[4] Reilly *et al.*^[1] also found that intussusception was far more common among children with untreated celiac disease than in the general pediatric population. Among 254 children with celiac disease, 1.2% experienced the intussusception <9 months before their diagnosis with celiac disease compared with 0.07% of children of their institution in the same time period. They concluded that the diagnosis of celiac disease should be considered in children with intussusception, even in the absence of growth failure.

Borkar VV *et al.* has stated that intussusception is frequently (25%) seen in children with newly diagnosed celiac disease, generally asymptomatic and resolves spontaneously on gluten free diet. It is often associated with more severe disease.^[5]

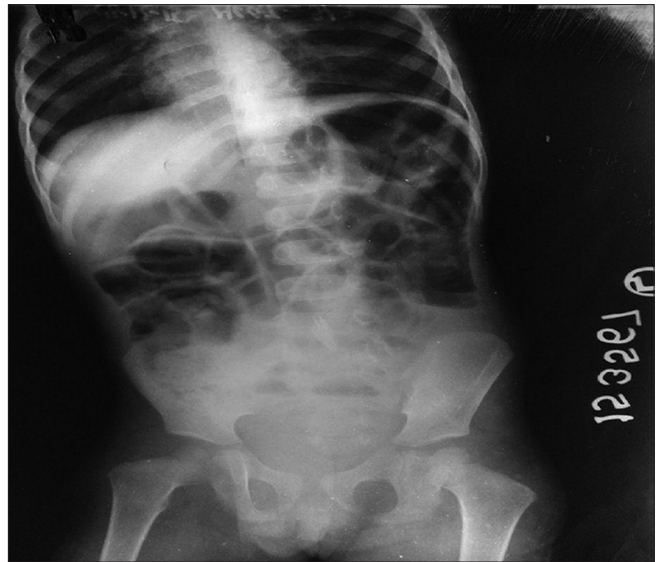


Figure 1: X-ray abdomen erect showing multiple abnormal air-fluid levels and gas-filled bowel loops in the abdomen

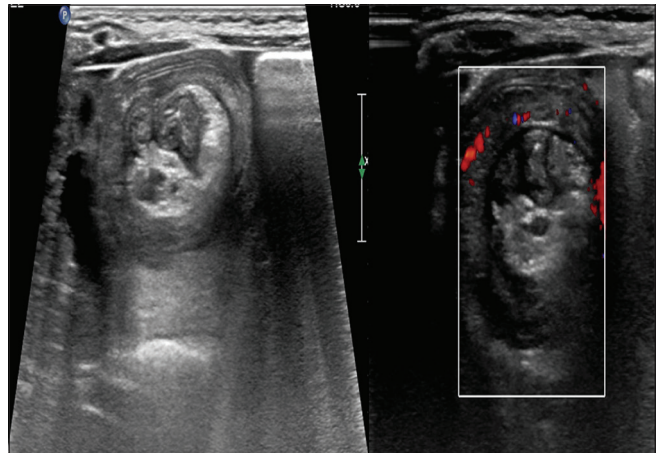


Figure 2: Ultrasound showing well-defined round to oval heterogeneously hypoechoic lesion with internal vascularity giving typical bowel within bowel appearance (Target sign) suggestive of intussusception

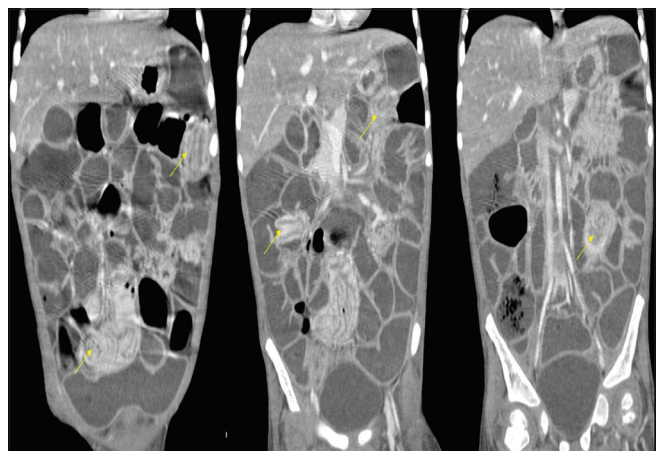


Figure 3: CT abdomen with contrast reformed coronal images showing telescoping of proximal bowel loops into distal loops at five different sites corresponding with multiple intussusceptions

Conclusion

Workup for diagnosis of celiac disease should be considered in children whose initial ultrasound or CT scan reveals intussusception especially in the presence of growth failure. So that unnecessary surgical intervention for the treatment of intussusception can be prevented.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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