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Case Report

Abdominal intussusception associated with coeliac disease: case report and literature review

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ABSTRACT

In genetically susceptible individuals, gluten ingestion triggers and immune infiltration and bowel damage in the classical pattern of coeliac disease, with variable symptoms. Intussusception is a condition where one segment of intestine 'telescopes' inside of another portion of intestine, which may cause symptoms of abdominal pain due to obstruction. Intussusception has been associated with coeliac disease. We report a 4-year-old girl presented with recurrent abdominal pain of variable severity and found to have intussusception on two occasions, which on both occasions reduced spontaneously during ultrasound examinations. She was later diagnosed with coeliac disease. This case highlights the importance of considering coeliac screening in patients with a history of recurrent abdominal pain and intussusception.

Keywords: Intsussussception, Coeliac disease, Abdominal pain

INTRODUCTION

Intussusception (IS) in children can present with classical symptoms of acute abdominal pain, red currant jelly stools and an abdominal mass. However, the clinical presentation varies and sometimes the patients can be asymptomatic, or the symptoms can be transient, presenting as recurrent abdominal pains with variable intensity.[1] IS may occur at any age but is commonly seen in children between 2-month to 2 years age group, with a peak incidence at the age of 5-9 months. It is considered to be an idiopathic in approximately 75% of affected children, lymphoid hyperplasia of the small bowel being the likely lead point. Other reported neurological factors are, tumors, Mickels' diverticula or associated condition such as Henoch-Schonlein purpura, viral infections and past versions of oral rotavirus vaccine.[2-4] Patients with Crohn's disease have been reported to be at an increased risk of developing intussusception.[5] Holsclaw et al in 1971 described 1% prevalence of IS among 2200 patents with cystic fibrosis.[6] Coeliac disease (CD) is an immune mediated enteropathy induced in genetically susceptible individuals after ingestion of gluten protein found in wheat, rye and other grains. CD is characterized by CD-specific antibodies and enteropathy with variable clinical manifestations. IS may or may not require treatment depending on the severity and the underlying condition.

CASE REPORT

A 4-year-old girl presented to our primary care clinic with intermittent episodes of colicky abdominal pain for 2 weeks. The severity of pain was described to vary between mild to very severe with no change in bowel habits. Each episode lasted for 10-15 minutes. She reported being completely asymptomatic between the intermittent bouts of abdominal symptoms. She reported an increase in the frequency of abdominal pains, but denied any infective

symptoms, vomiting or skin rashes. She had no history of any abdominal surgeries or documented food or medication allergies.

On physical examination, she was pain free. Her vital signs were normal. Growth normal with height percent weight percent, an abdominal examination was consistent with soft non-tender abdomen without masses or hepato-splenomegaly, although later she had tenderness in the upper abdomen.

To rule out an intra-abdominal pathology for her ongoing intermitted episodic distressing abdominal pain, an urgent abdominal ultrasonography was performed which revealed a few reactive lymph nodes in the mesentery of the bowel and a segment of the small bowel intussusception extending over 70 mm. Vascularity was seen in all 3 layers of the bowel. She was admitted to the hospital and underwent repeated ultrasound examinations on her abdomen confirming that the IS had self-resolved without any surgical interventions. A month later, she re-admitted to the emergency department with similar symptoms of abdominal pain, and her abdominal ultrasonography examination confirmed small bowel intussusception with a target sign measuring 22×15×27 mm (Figure 1).

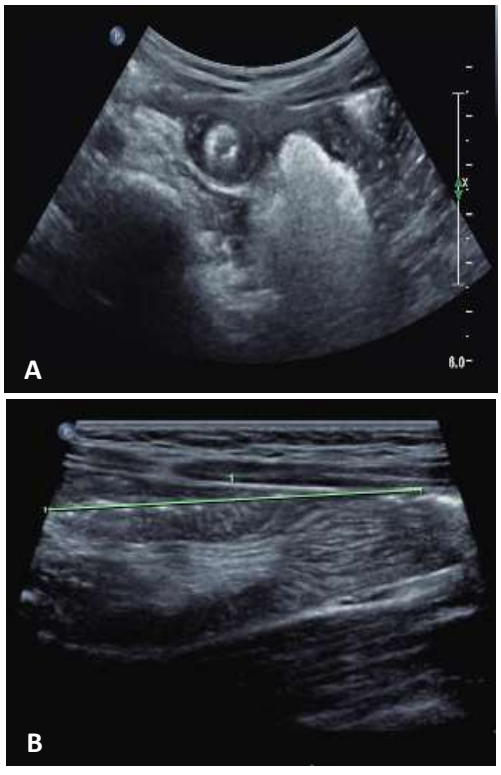

Figure 1: Ultrasound showing intussusception of small bowel (A) transverse axis; (B) longitudinal axis.

In addition, multiple lymph nodes were seen in the left upper quadrant and small amount of fee fluid in the right iliac fossa. The intussusception completely reduced spontaneously during the examination. She was discharged from the hospital with a plan to follow up as an out-patient with coeliac bloods suggested if symptoms persisted. Following discharge, she continued to have abdominal pain intermittently, so she was screened for CD.

Her anti-tissue transglutaminase tTG (IgA) was significantly elevated (>1000 CU). An Upper GI Endoscopy and the microscopic findings on duodenal biopsy were consistent with CD (Figure 2). Since commencing gluten-free diet, no further episodes of IS were evident. Laboratory studies performed confirmed compliance with gluten free diet with expected decline from in tTG antibodies from 1000 CU to 19 CU (negative<20 CU) by 11 months post diagnosis.

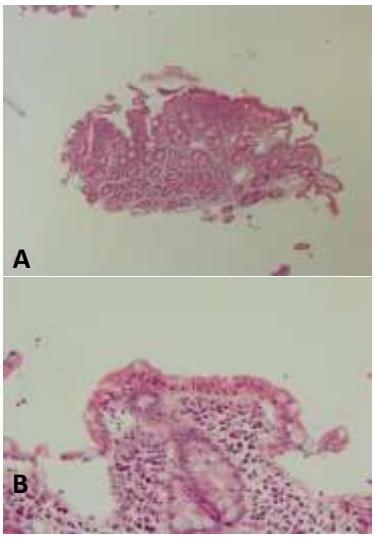

Figure 2: Histopathology shows (A) villous atrophy and expansion of the lamina propria with chronic inflammatory cell infiltrate at 4X magnification; (B) intraepithelial lymphocytosis at 20X magnification.

DISCUSSION

It is the most common abdominal emergency affecting children, particularly younger than two years of age.[7] IS has been rarely reported in children as a presenting symptom of CD. [8]The reported prevalence of IS with CD is around 1.6-20% and 1.2-25% in adults and children respectively, but these figures could be higher because of the episodic presentation.[8-11]

Association of IS and CD for the first time was demonstrated by Ruoff M et al in 1968 in adults and by Germann R et al in children in 1997.[12-14]

Rilley et al, found that the frequency of IS was greater in the children with untreated CD than in the general paediatric population.[8] In their study of 254 children with CD,1.2% experienced the IS in the 9 months before their diagnosis with CD compared to 0.07% of the children of their institution in the same time period. The majority of these children with CD who were found to have IS had no nutritional deficiency evidenced at the time diagnosis of the IS. On the basis of their study, they concluded that CD

should be considered as the underlying cause in children presenting with IS, even without nutrients depletion. Gheibi S et al found that 5% of the paediatric referrals to the gastrointestinal ward with CD had IS.[9] In this prospective study of 150 children with CD on the basis of their positive antibodies and duodenal biopsy, with median age of 72 months (range: 16-204) were screened, irrespective of their symptoms, 37 out of 45 children (25%) were found to have ID with 95% involving the small bowel. All but one had asymptomatic Intussusception. On initiation of the gluten-free diet, IS resolved spontaneously in 65% children within 7 days, 84% within 14 days and 92% within 28 days. No patients required surgical or hydrostatic reduction. This study has shown that children with IS were more likely to have diarrhoea, rickets, oedema and refeeding syndrome.[14]

Gonda TA et al determined the prevalence of IS among a cohort of 880 patients with CD (age 47±17.5 years).[10] Of these patients,14 had IS diagnosed, among which 3 were found incidentally whilst eleven of them had abdominal pain. IS was the initial manifestation of CD in 57% (8/14) and was associated with abdominal pain. In majority of patients, symptoms resolved following adherence to a gluten-free diet.

In contrast Ludvigsson et al in a large case-control study from Sweden evaluated the risk of late CD in patients with IS, which studied 144,552 individuals (40.7% were ≤19 years of age) and 29,096 CD patients (40.6% were ≤19 years of age), IS was not a risk factor for later CD [34 cases of IS in CD (0.12%) vs 143 (0.10%) cases in controls].[15] They did not find any significant association between IS and future CD, but noted a modest increased risk of IS after diagnosis of CD. Taken together this evidence suggests that IS in children around the time of CD diagnosis is likely a function of the immune activation and lymphoid hyperplasia associated with its pathogenesis.

CONCLUSION

Our patient's presentation and the literature review suggest that children presenting with recurrent abdominal pain and an intussusception episode (particularly outside the typical age-group) should be considered for CD screening at presentation.

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