Case and Review

Transient Small-Bowel Intussusception Unmasking Adult Celiac Disease

Maryam AlAhmad^a Abdulqader Almessabi^b

^aGastroenterology Fellow, Sheikh Shakhboub Medical City, Abu Dhabi, UAE;

^bGastroenterology Consultant, Sheikh Shakhboub Medical City, Abu Dhabi, UAE

Keywords

Celiac disease · Intussusception · Tissue transglutaminase antibodies · Diarrhea · Abdominal pain

Abstract

Celiac disease (CD) is a chronic immune-mediated small-intestinal disorder. The tissue transglutaminase (TTG) antibodies test is the initial step in the diagnosis of CD. Intussusception is a process where one segment of the bowel invaginates into the lumen of an adjacent segment. Despite that, the association between adult intussusception and CD has been described previously, and it is still not widely recognized. We report a case of adult intussusception as the first manifestation of CD. A 28-year-old nonalcoholic and nonsmoker, previously healthy man, presented to the emergency department with a 1-week history of progressive diffuse abdominal pain associated with abdominal distension, nausea, vomiting, diarrhea, and 7 kg weight loss. The physical exam was remarkable for thin body habitus with no lymphadenopathy. Laboratory testing was significant for hypochromic microcytic anemia, AST 214 IU/L, ALT 217 IU/L, alkaline phosphatase 183 IU/L, and INR 2.9. An abdominal radiograph showed distended gas-filled small- and large-bowel loops. An abdominal computed tomography scan with contrast was done and demonstrated a telescoping of the small bowel in at least 3 areas with a target sign appearance with no signs of obstruction that suggested small-bowel intussusception. Following this imaging result, the patient was admitted under general surgery and kept nothing by mouth. The surgical team approached the gastroenterology team for push enteroscopy with biopsy which the patient refused. Further investigation included TTG antibodies; IgA and IgG were significantly elevated, 4,965.5 CU and 431.9 CU, respectively (reference: <19.9 CU). The diagnosis of CD leading to adult intussusception was made. The patient

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Correspondence to:
Maryam AlAhmad, maryam.alahmed@live.com

was advised to start a gluten-free diet. During the hospital stay, the patient had complete resolution of his symptoms and was discharged home. In summary, this case highlights the potential link between adult intussusception and CD. Adult intussusception is a rare condition that its underlying etiology should be meticulously investigated to expedite treatment and prevent unnecessary surgical intervention.

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Introduction

Celiac disease (CD) is a chronic immune-mediated small-intestinal disorder triggered by gluten ingestion elicited in individuals carrying HLA-DQ2 or HLA-DQ8 genotypes [1]. Classical manifestations include diarrhea, abdominal pain, and weight loss. However, CD can present with anemia, osteoporosis, or arthralgia. Intussusception is a process where one segment of bowel invaginates into the lumen of an adjacent segment. Intussusception is a rare clinical condition in adults [2]. Despite that, the association between intussusception and CD has been described previously, and it is still not widely recognized. Here, we report a case of adult intussusception as the first manifestation of CD.

The Case History

A 28-year-old, nonalcoholic and nonsmoker, previously healthy man presented to the emergency department with 1 week history of diffuse intermittent dull abdominal pain which was gradually and progressively exacerbated by food intake, especially wheat products, with no relieving factors associated with abdominal distension, nausea, vomiting, diarrhea, and 7 kg weight loss. He denied fever, night sweat, passage of fresh blood per rectum, having blackish tarry stool, or yellowish discoloration of the sclera. No history of skin rash or joint pain was also observed. He had no history of similar attack before and no significant family history. In addition, surgery or drug history was negative. He came back from Pakistan 2 months ago.

The physical exam was remarkable for thin body habitus and no lymphadenopathy, with normal bowel sounds and a soft nontender abdomen. No organomegaly could be observed. A digital rectal exam showed no rectal masses, hemorrhoids, or gross blood.

Laboratory testing was significant for hypochromic microcytic anemia, abnormal liver function (AST 214 IU/L [reference: <50 IU/L], ALT 217 IU/L [reference: <40 IU/L], and alkaline phosphatase 183 IU/L [reference: 40–129 U/L]), with a normal total bilirubin level and INR 2.9. Investigation of the elevated liver enzymes revealed negative autoimmune and viral serology. His workup for infectious causes of diarrhea was negative. An abdominal radiograph showed distended gas-filled small- and large-bowel loops. An abdominal sonogram revealed a normal liver with patent vasculature, an evidence of thickened loop of bowel with features suggestive of intussusception with a fluid-filled dilated bowel.

An abdominal computed tomography (CT) scan with IV contrast was done and demonstrated a telescoping of the small bowel in at least 3 areas with a target sign appearance (shown in Fig. 1, 2) with no free fluid presented or enlarged lymph nodes and no signs of obstruction suggested of small bowel intussusception. Following this imaging result, the patient was admitted under general surgery and kept nothing by mouth (NPO). The surgical team approached the gastroenterology team for push enteroscopy with biopsy which the patient refused.

Fig. 1. Abdominal CT scan with IV contrast, axial section showing a multiple telescoping appearance of the small bowel with a classical target sign (white arrows).

Fig. 2. Abdominal CT scan with IV contrast, sagittal section showing invaginated 224 mesenteric fat and vessels (white arrows).

Further investigation included (tissue transglutaminase [TTG]) antibodies IgA and IgG being significantly elevated, 4,965.5 CU and 431.9 CU, respectively (reference: <19.9 CU). The diagnosis of CD leading to adult intussusception was made.

The patient was advised to start a gluten-free diet. During the hospital stay, the patient had complete resolution of his symptoms and was discharged home. The patient traveled back to his home country for further evaluation.

Discussion

Adult intussusception remains a rare condition. It can be classified by underlying etiology as primary or idiopathic that occurs in the absence of a lead point, whereas in the secondary, a lead point is identified [2]. In a cohort of 58 surgically proven adult intussusception cases, 90% of cases were thought to be secondary to structural lesions [3]. CD stands out as least

likely to be associated with intussusception. We are reporting a case of transient small-bowel intussusception, revealing CD in early adulthood life.

The association between CD and idiopathic intussusception has first been demonstrated in 1968 by Ruoff et al. [4]. Theories of adult intussusception development in CD are thought to invagination of one segment bowel loop into another due to dilated flaccid loops, disturbing normal peristaltic waves [5]. Sanders et al. [6] found that 4.9 years is the mean time from the symptoms onset to diagnosis of CD.

Nausea, vomiting, change in bowel habit, and abdominal distension are all features of intussusception in adults. Nowadays, intussusception is diagnosed by CT. The classic CT scan finding is the “target” sign or “sausage-shaped” soft-tissue mass [7]. In a case series involving 28 CT scans of CD adult patients who were reviewed, Tomei et al. [8] reported that intussusception was observed in up to 21%. However, other CT scan findings have been reported in CD such as small-bowel folds pattern abnormalities, intestinal dilatation, bowel wall thickness, and lymphadenopathy [8].

Different etiological factors play a role in adult intussusception. Among them are organic lesions such as inflammatory bowel disease, Meckel’s diverticulum, benign or malignant neoplastic lesions, and iatrogenic or less commonly occurring CD [9]. Clinical, serological, and histological features are needed to diagnose CD. Anti-TTG sensitivity and specificity are 98.6 and 91%, respectively [10].

Our patient presented with nonspecific gastrointestinal symptoms followed by CT scan imaging, confirming the diagnosis of adult intussusception with significantly elevated anti-TTG. In view of patient preference, endoscopic evaluation was not performed. Gluten-free diet was initiated, and the patient improved conservatively without the need for surgical intervention.

In conclusion, this case highlights the potential link between adult intussusception and CD. Adult intussusception is a rare condition that its underlying etiology should be meticulously investigated to expedite treatment and prevent unnecessary surgical intervention.

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Statement of Ethics

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. Ethics approval for case report was not required. This has been approved by the Research and Ethics Committee in Sheikh Shakhbout Medical City.

Conflict of Interest Statement

The authors declare that they have no conflicts of interest.

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Author Contributions

M.A. wrote the case and reviewed the literature. A.A. reviewed the manuscript and revised it.

Data Availability Statement

The data used to support this case is available within the article. Further inquiries can be directed to the corresponding author.

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