Celiac disease presenting with intestinal obstruction: Report of two cases

İntestinal obstrüksiyon ile prezente olan çölyak hastalığı: İki vaka sunumu

To the Editor,

The presentation of celiac disease (CD) has changed over time, and classical gastrointestinal manifestations as the presenting features are less common. Gastrointestinal features such as abdominal pain and constipation and non-gastrointestinal findings, such as short stature, delayed puberty and iron deficiency anemia, are frequently encountered as presenting symptoms. Here, we report two pediatric patients with CD presenting with small bowel intussusception and pseudoobstruction, respectively.

A 20-month-old girl was referred to our hospital due to weight loss, vomiting and diarrhea, which began approximately three months before. Two weeks before her admission, she was hospitalized because of abdominal distention and severe vomiting. Abdominal ultrasonography revealed an intussuscepted bowel segment with proximal dilatation and loss of motility at the distal segments. Intussusception was resolved by barium enema. She was referred to our hospital because of ongoing diarrhea and vomiting after resolution of the intussusception. Laboratory tests showed antigliadin immunoglobulin (Ig) A (AGA IgA) >100 U/ml (normal range: 0-12), antigliadin IgG (AGA IgG) >100 U/ml, tissue transglutaminase IgA (tTG IgA) > 200 U/m, and strong (++++) positive anti-endomysial antibody (EMA). Duodenum biopsy revealed Marsh type 3a mucosa, and the diagnosis of CD was confirmed. After 10 months of a strict glutenfree diet (GFD), she gained nearly 5 kg, and the results for antigliadin, tissue transglutaminase and anti-endomysial antibodies were negative. She also had no more vomiting or diarrhea.

Another patient, a 2.5-year-old girl, was evaluated for constipation, which began at the age of three months. One month before her admission, she was hospitalized because of abdominal pain, distention and vomiting. Her abdominal X-ray revealed air-

fluid levels, indicative of bowel obstruction (Figure 1). There was no sign of intussusception on ultrasonographic examination. She was referred to our hospital after being evaluated thoroughly for the cause of obstruction, including a rectal suction biopsy for Hirschsprung's disease, which was found to be negative. Laboratory tests revealed anemia (hemoglobin: 10.4 g/dl), normal levels of serum electrolytes, AGA IgA >100 U/ml, AGA IgG >100 U/ml, tTG IgA >200 U/ml, and positive (++++) EMA. Diagnosis of CD was confirmed with endoscopic duodenum biopsy. After GFD of three months, she gained 1.5 kg and her constipation was resolved.



Figure 1. Air-fluid levels in abdominal plain X-ray of Patient 2.

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Small bowel intussusception has been known to be associated with CD. In the past decades when barium radiologic studies were more commonly used in the diagnosis of CD, transient intussusceptions were seen in 20% of adults and children with proven CD, mostly asymptomatic (1). Intussusception was also recognized as a presenting symptom of pediatric CD (2), and recurrent intussusceptions were also reported (3). There is usually no identifiable lead point (4), and intussusception resolves without surgical intervention.

Intestinal pseudoobstruction is the condition in which there is no mechanical cause to explain the obstruction. It is a rare complication of CD and is usually reported in adults (5). As in intussusception, the mechanism of pseudoobstruction in CD is

not fully established. Gastrointestinal motility disorders in CD were proven previously. The mechanism for motility disturbances can originate from reduced absorption of food constituents, immunologic reactions or hormonal derangements (6). It has been suggested that intestinal motor abnormalities create hypotonic flaccid bowel loops, which disturb normal peristalsis and cause intussusception without a lead point (7). Delayed short bowel transit time and reduced motility can be responsible for the signs and symptoms of mechanical or pseudoobstruction in children with CD.

In conclusion, CD should be kept in mind in children with symptoms of small bowel intussusception or pseudoobstruction without an underlying

REFERENCES

- 1. Cohen MD, Lintott DJ. Transient small bowel intussusception in adult celiac disease. Clin Radiol 1978; 29: 529-34.
- 2. Lastennet F, Piloquet H, Camby C, et al. Acute intestinal invagination revealing celiac disease in a 9-month-old infant. Arch Pediatr 2002; 9: 151-4.
- 3. Altaf MA, Grunow JE. Atypical presentations of celiac disease: recurrent intussusception and pneumatosis intestinalis. Clin Pediatr (Phila) 2008; 47: 289-92.
- 4. Buckley O, Brien JO, Ward E, et al. The imaging of coeliac disease and its complications. Eur J Radiol 2008; 65: 483-90.
- 5. Koklu S, Coban S, Ertugrul I, et al. Intestinal obstruction in celiac disease: case report. Dig Dis Sci 2004; 49: 1485-8.
- Tursi A. Gastrointestinal motility disturbances in celiac disease. J Clin Gastroenterol 2004; 38: 642-5.
- Basotti G, Castellucci G, Betti C. Abnormal gastrointestinal motility in patients with celiac sprue. Dig Dis Sci 1994; 39: 1947-54.

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Pulse granuloma, unusual localization: appendix

Puls granülom, farklı lokalizasyon: apendiks

To the Editor,

A 37-year-old male patient referred to the emergency service of the hospital with the complaints of abdominal pain and nausea for two days. Abdominal ultrasonography displayed non-compressed appendix vermiformis consistent with appendicitis.

In the surgical exploration, a retrocecally localized inflamed and edematous appendix vermiformis was detected.

Macroscopic examination showed a large fattycoated appendix with obliterated lumen.

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