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Original Article

Is it necessary to screen for celiac disease in all children with intussusception?

Melike Arslan[1], Gökhan Berktuğ Bahadır[2], Mehmet Bahadır Çalışkan[2], Coşkun Fırat Özkeçeci[1], Edibe Gözde Başaran[1], Sevim Ecem Ünlü Ballı[2], Hüseyin Emre Atasever[2], Necati Balamtekin[1]

[1]Division of Pediatric Gastroenterology, Department of Child Health and Diseases, Gülhane Training and Research Hospital, Ankara, Türkiye
[2]Department of Pediatric Surgery, Gülhane Training and Research Hospital, Ankara, Türkiye

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ABSTRACT

Objective: While there is a close relationship between celiac disease (CD) and intussusception, it is not yet clear whether the detection of intussusception in children requires physicians to always screen for CD. In our study, we aimed to determine the frequency of CD in children with intussusception and to find the answer to whether CD screening is necessary for every child with intussusception.

Methods: The study included 50 symptomatic pediatric patients diagnosed with intussusception who were followed up and treated in the Health Sciences University Gülhane Training and Research Hospital Pediatric Surgery Clinic between 2020–2023. CD screening was performed in patients followed up with a diagnosis of intussusception.

Results: The mean age of the patients was 3.61 ± 2.02 years, and 33 (66%) were male. Of 62 intussusceptions observed in 50 patients, 46.8% were ileo-ileal, 35.5% ileo-colic, 11.3% jejeno-jejunal, 4.8% colo-colic, and 1.6% recto-sigmoid. CD was diagnosed endoscopically in three (6%) patients with an intussusception diagnosis and an anti-tissue transglutaminase level > 200 IU/ml. Intussusceptions in 43.5% of the patients were reduced by hydrostatic reduction, 41.9% spontaneously, 9.7% laparotomically, and 4.8% laparoscopically. A single intussusception attack occurred in 82% of patients; 14% had two, 2% had three, and 2% had four. No significant correlation was found between the number of intussusception attacks and the presence of CD (p = 0.34). There was also no relationship between the type of intussusception and age (p = 0.74), gender (p = 0.24), or treatment (p = 0.12) or between the presence of CD and gender (p = 0.26), age (p = 0.68), or type (p = 0.28) of intussusception.

Conclusions: CD is more common in symptomatic children with intussusception than in healthy children. Screening pediatric patients with idiopathic intussusception for CD may reduce the recurrence of intussusception and complications and morbidities that may occur due to a delayed CD diagnosis.

Keywords: Celiac Disease, intussusception, screen, children

INTRODUCTION

Celiac disease (CD) is a chronic autoimmune disease that destroys the intestinal mucosa due to a series of abnormal immune responses triggered by gluten intake in susceptible individuals.[1] Frequency studies show that the disease affects approximately 1% of the world's population and our country (Turkey).[2,3]

According to CD presentation, it can fall into one of three categories: the typical form (characterized by abdominal distention, chronic diarrhea, and failure to thrive), the atypical

Correspondence: Melike Arslan **E-mail:** melikearslan190@gmail.com
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form (characterized by isolated short stature, delayed puberty, refractory iron deficiency anemia, chronic constipation, abdominal pain, aphthous stomatitis, enamel defects, and osteoporosis), or silent disease.[4,5]

Intussusception is a form of intestinal obstruction in which one part of the intestine telescopes inside of another. It is the most common cause of gastrointestinal obstruction in children three months to 5 years of age.[6] Rarely, lead points such as Meckel's diverticulum or lymphoma may be found, but approximately 90%–95% of pediatric cases are idiopathic. It may be associated with those of known etiology, Henoch Schönlein purpura, polyps, duplication cysts, viral infections, cystic fibrosis, taking old versions of the oral rotavirus vaccine, Crohn's disease, and CD.[6-9]

Although it is not common, there are reports in the literature that celiac patients may present with intussusception as an atypical presentation finding.[6,10] The general prevalence of symptomatic intussusception in adults with CD is 1.6% and 1.2% in children.[6] Whether the detection of intussusception in children requires CD investigation still needs to be determined.

Thus, this study aimed to investigate the frequency of CD in children with intussusception and whether screening for CD in every child with intussusception is necessary.

MATERIALS AND METHODS

This study included 50 symptomatic pediatric patients diagnosed with intussusception by abdominal ultrasonography who were followed up and treated in the Pediatric Surgery Clinic of Health Sciences University Gülhane Training and Research Hospital between 2020–2023. Written consent was obtained from the patient's families to participate in the study, and approval was received from the local ethics committee with the date and decision number 2020-101/10.03.2020.

For CD screening in patients followed and treated for intussusception, serum IgA levels were measured using Beckman Coulter kits (Brea, CA), and anti-tissue transglutaminase IgA antibody levels were measured using ELISA kits (Orgentec, Germany). A pediatric gastroenterologist performed an upper gastrointestinal endoscopy to confirm the diagnosis of CD in three children with serum anti-tissue transglutaminase levels >200 IU/ml. Upper gastrointestinal endoscopy was performed with an Olympus X260 scope (Olympus Optical Corporation, Japan). One biopsy specimen from the duodenal bulb and four from the second part of the duodenum were obtained from each patient who underwent an endoscopy to diagnose CD histopathologically according to the Marsh classification system.[11] The hospital data recording system obtained the clinical findings, laboratory results, imaging, endoscopy, and histopathology results.

Statistical analysis

Data were evaluated using the Statistical Package for the Social Sciences for Windows version 20.0. Descriptive statistics were presented as numbers and percentages, mean ± standard deviation, minimum-maximum values, and median values for numerical variables. The conformity to the normal distribution was examined with the Shapiro-Wilk test, and the median difference between the groups was analyzed with the Mann-Whitney U, and Kruskal-Wallis H tests when the numerical variables did not show a normal distribution. Analyzing categorical variables among themselves was performed with the Chi-Square test or Fisher's Exact test. The tests were considered significant if the p-value was < 0.05, examining the data at the 95% confidence level. The tests were considered significant if the p-value was < 0.05, examining the data at the 95% confidence level.

RESULTS

The mean age of the patients was 3.61 ± 2.02 years, and 33 (66%) were male. A total of 62 intussusceptions were observed in 50 patients: 46.8% ileo-ileal, 35.5% ileo-colic, 11.3% jejeno-jejunal, 4.8% colo-colic, and 1.6% recto-sigmoid. CD was diagnosed endoscopically in three (6%) patients who underwent intussusception and had an anti-tissue transglutaminase level >200 IU/ml. The clinical features of the three patients diagnosed with CD are shown in Table 1. The frequency of intussusception was 2.1% (3/139) in 139 celiac patients followed up in our center. Intussusceptions were reduced in 43.5% of cases by hydrostatic reduction, 41.9% spontaneously, 9.7% laparotomically, and 4.8% laparoscopically. A single intussusception attack occurred in 82% of patients, two in 14%, three in 2%, and four in 2%. No significant correlation was found between the number of intussusception attacks and the presence of CD ($p = 0.34$). There was no relationship between the type of intussusception and age ($p = 0.74$), gender ($p = 0.24$), or treatment ($p = 0.12$). There was also no relationship between the presence of CD and gender ($p = 0.26$), age ($p = 0.68$), or type ($p = 0.28$) of intussusception.

DISCUSSION

Although intestinal intussusception is generally seen in children between the ages of 3 months and 3 years, the highest incidence is seen between 4 and 9 months[12], and it is detected

Table 1. Clinical characteristics of those diagnosed with CD among intussusception patients						
Patient	Age of Intussusception/ CD diagnosis	Intussusception type	Number of intussusception attacks	Treatment	Histological classification	Follow-up
Patient 1	5 y 6 mo/ 5 y 7 mo	Jejeno-jejunal	1	hydrostatic reduction	Marsh 3A	Gluten-free diet, clinically good, 7 y 1 mo
Patient 2	5 y/5 y 1 mo	Ileo-colic	1	hydrostatic reduction	Marsh 2	Gluten-free diet, clinically good, 6 y 6 mo
Patient 3	17 mo/19 mo	Ileo-colic Ileo-ileal Ileo-colic Ileo-colic	4	hydrostatic reduction	Marsh 3B	Gluten-free diet, clinically good, 4 y 10 mo

approximately twice as often in boys than in girls.[13,14] Similar to the literature, the mean age of the patients in our study was 3.61 ± 2.02 years, and 66% were male.

Pediatric intussusceptions are divided into six types: ileo-colic, ileo-ileocolic, colo-colic, ileo-colocolic, ileo-ileal, and jejuno-jejunal. The ileo-colic type, in which the distal ileum invades the cecum through the ileocecal valve, is the most common type of intussusception, constituting 90% of cases.[12,13] Intussusception often recurs regardless of the reduction method. Some studies have shown that approximately 10% (8-15%) of patients experience recurrent intussusception.[15] In our study, recurrent intussusception was observed at a rate of 18%, and of all intussusceptions, 46.8% were ileo-ileal, 35.5% ileo-colic, 11.3% jejeno-jejunal, 4.8% colo-colic, and 1.6% recto-sigmoid.

The preferred initial treatment method in most cases of intussusception is an image-guided reduction with pneumatic or hydrostatic enema.[16] Conditions requiring surgical intervention include peritonitis, shock, sepsis, perforation, recurrent enema failure, and persistent symptomatic small bowel obstruction.[17] In our study, 43.5% were reduced by hydrostatic reduction, 41.9% spontaneously, 9.7% laparotomically, and 4.8% laparoscopically. Small bowel intussusception (ileo-ileal and jejuno-jejunal) accounted for 88.4% of patients with spontaneous reduction.

Intussusception may occur in patients with CD, but it is mostly asymptomatic. Controversy continues regarding whether all children with idiopathic intussusception should be screened for CD to prevent future episodes of obstruction.[18] Because CD is a chronic inflammatory condition, the suggested cause of intussusception in CD cases is inflammation and thickening of the intestinal wall, leading to hyperperistalsis and subsequent enlargement of the small intestine. One intestinal loop segment is thought to be invaginated into the other due to dilated drooping rings that disrupt normal peristaltic waves.[18,19]

Borkar et al. investigated the prevalence and natural history of intussusception in newly diagnosed CD prospectively and found subclinical intussusception in 25% of children with newly diagnosed CD.[10] Another study revealed that 1.2% of children with CD experienced an intussusception before treatment with a gluten-free diet.[6] Reilly et al. found a higher incidence of intussusception among children with CD compared to the general pediatric population in their study, and they argued that the underlying cause of intussusception in children might be related to CD, even if the patient appears well.[6] The reported prevalence of intussusception in adult patients with CD is approximately 1.6% to 20%.[20] In a study of American adults, intussusception was identified as the first sign of celiac disease in 57% of cases. Paul et al. suggested that while radiologists investigate intussusception with ultrasonography, they can also evaluate key features of CD, such as increased bowel wall thickness, free peritoneal fluid, and enlarged mesenteric lymph nodes.[21] The frequency of intussusception was 2.1% (3/139) in 139 celiac patients followed up in our center.

In a large population-based case-control study on the relationship between intussusception and CD, it was found that there was no relationship between intussusception and future CD. However, there was a 2-fold increase in intussusception risk after a CD diagnosis.[22] Aldaher et al.[23] found that the frequency of CD in children with intussusception was as high as 10.5% (6/57). In our study, CD was diagnosed endoscopically in 3 (6%) of 50 symptomatic pediatric patients with intussusception. One of the patients diagnosed with CD had recurrent intussusception with four attacks in the two months before diagnosis. There was no significant relationship between the number of intussusception attacks and the presence of CD or the presence of CD and gender, age, or type of intussusception in our study. While we did not include a control population of healthy children to see the prevalence of CD in healthy children of the same age in our community, in a study of 20,190 healthy children in Turkey, the

prevalence of CD was 0.47%, and the global prevalence of CD was approximately 1%.[2,3] The frequency of CD in our symptomatic patient population with intussusception (6%) was significantly higher than that of the healthy population.

In conclusion, CD was seen more frequently in symptomatic children who have undergone intussusception than in normal healthy children. Our study's important limitations are that it was single-center and conducted with a limited number of patients. Screening pediatric patients with idiopathic intussusception for CD may reduce the recurrence of intussusception and complications and morbidities that may occur due to a delayed CD diagnosis.

Ethical approval

The study was approved by the local ethics committee (2020-101/10.03.2020).

Author contribution

Study conception and design: MA, GBB, NB; data collection: MA, CFÖ, EGB, SEÜB, HEA; analysis and interpretation of results: MA, GBB, MBÇ; draft manuscript preparation: MA. All authors reviewed the results and approved the final version of the manuscript.

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Conflict of interest

The authors declare that there is no conflict of interest.

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