# CASE REPORT

# Simultaneous Intussusception in Monozygotic Twins Monozigot İkizlerde Eşzamanlı İnvajinasyon

Abstract

of the disease.

Öz

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Intussusception is a well-known disease, and one of the most frequent causes of acute bowel obstruction in children aged 3-18 months, with an incidence of 0.1-0.4% of live births. Although idiopathic intussusception has not been considered a genetic disease, it has also been reported in families and relatives (1-4). Herein, we present the first case report in the English literature of simultaneous occurring intussusception in monozygotic twins.

In this case report, it was aimed to present the simultaneously occurring intussusception in the monozygotic twins. In addition to genetic predisposition, environmental factors has been hypothesized to be responsible for the development

Bu olgu sunumunda monozigot ikizlerde aynı anda ortaya çıkan invajinasyon sunulmuştur. Genetik predispozisyona ilaveten, çevresel etkenlerin de hastalığın

gelişiminden sorumlu olabileceğini hipotezi ileri sürülmektedir.

# **Case Report**

Introduction

A previously healthy 6-month-old boy who one of monozygotic twins, presented at the pediatrics department with a 1-day history of nonbilious vomiting and diarrhea, and had hospitalized to the department of pediatrics with diagnosis of acute gastroenteritis. The clinical symptoms had worsened progressively at the morning with an increase in bilious vomiting and a decrease in stooling. On examination, no abdominal distention was found, but a nontender mass palpated in the right upper quadrant of abdomen, and a rectal examination revealed the rectum contained of mucous and bloody stool.

The twin of the first infant admitted to our surgical department with similar complaints on the operation day of his brother. A more detailed history revealed that the complaints started the same day with his brother, and they admitted to hospital with together two days ago. The abdomen was soft and non-distended with a palpable mass in the right upper quadrant. There were no peritoneal signs, and rectal examination was revealed bloody stool. In both of them, the abdominal US showed a characteristic target sign in right upper abdomen.

## Keywords

Intussusception, twin, genetic, environmental factors

#### Anahtar kelimeler

İnvajinasyon, ikiz, genetik, çevresel faktörler

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© The Journal of Current Pediatrics, published by Galenos Publishing. © Güncel Pediatri Dergisi, Galenos Yayınevi tarafından basılmıştır. Hydrostatic reductions with contrast enema was attempted, but unfortunately was unsuccessful. The following same operative findings were observed in both cases: an ileocolic intussusception, reactive mesenteric lymphadenopathy, thickened and mildly ischemic patch (Peyer's patch) on the anti-mesenteric side of terminal ileum, and mobile cecum. The intussusceptions were reduced and appendectomy was performed. Rotavirus stool antigen was negative in the patients. The patient's postoperative courses were uneventful, and they were discharged on the fourth post-operative day. In the sixth month of the follow-up period, the twins were healthy (Figure 1).

# Discussion

Simultaneous intussusception occurring in twins has been previously reported (2,3). Thomas and Zachary (1) reported that intussusception diagnosed with an interval of 36 hours in a pair of 2-year-old identical twins. Nakanishi et al. (2) also reported 18-month-old dizygotic twins who simultaneously developed ileocecal intussusceptions preceded by generalized varicella infection. To our knowledge, this is the first report in English literature of intussusception occurring simultaneously in monozygotic twins.

The etiology of idiopathic intussusception is considered to be incidental, and it has not been traditionally regarded as having any hereditary basis. On the other hand, a genetic predisposition has been suggested in families (3). There was estimated that the risk to siblings is about 5-20 times more than that for the general population in literature (4).

Many hypotheses have been proposed to explain the etiology of idiopathic intussusception such as a swollen Peyer's patch as lead point due to either viral infections or an immunologic response, a disproportion between the size of the ileum and the ileocecal valve, and lack of normal rotation and fixation of the intestine (Waugh's syndrome) (3-5). Except first one, the possible causes of the intussusception in present cases were developmental. The above-mentioned etiological factors were observed in our cases. Although no viral or bacterial organism isolated from our patients, there was a history of acute gastroenteritis which started at the same time in twins.

Little is known of etiology of twin intussusception, however; most of the relevant data have been

obtained in the case reports, and are inappropriate for explanation of exact etiology. Thomas and Zachary (1) have suggested that the enlarged lymphoid tissue due to infection is an important etiological factor. Nakanishi et al. (2) put forward that anatomic enlargement itself may be the most important factor in predisposing to intussusception. Hsu et al. (6) have speculated that an anatomic predisposition to intussusception may exist and may be inherited in an unknown pattern favoring its occurrence in siblings. In addition, we hypothesized that similar response to same environmental factors may be inherited in monozygotic twins who predisposed to intussusception anatomically. Zhernakova et al. (7) indicated that genetic factors play an important part in the development of immune-related disorders, and the heritable component of these disorders is evident from the high concordance observed in monozygotic twin pairs and increased familial clustering. Bouma and Strober (8) have also reported that the host genetic background determines the susceptibility to intestinal inflammation. Thereby, the inherited inflammatory response due to same infection in twins may cause to hypertrophy in the Peyer's patches as same degree. However more studies are needed to prove the inherited etiology of idiopathic intussusception.

Although our cases are unique, the etiology of twin intussusception seems reasonable to assume that same as the other idiopathic familial and twin cases. According to literature and our experience, no single cause can account for the etiology of idiopathic intussusception in twins. Rather, it appears to be the result of multiple causes such as similar systemic



Figure 1. Postoperative picture of healthy twins

inflammatory response, and same congenital anatomic and genetic predispositions. In conclusion, same environmental factors at same time may cause to simultaneous onset intussusception in monozygotic twins.

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