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CASE REPORT

Sequential intussusception in a set of female twins

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Abstract

***Introduction*:** Intussusception is the leading cause of intestinal obstruction in children in sub-Saharan Africa. Familial occurrences of intussusception have been reported. Simultaneous or concurrent twin intussusception is rarely reported, particularly in Africa.

***Case presentation*:** We report a case of intussusception in 5-month-old dizygotic female twins occurring 18 days apart in Southern Nigeria. The younger twin presented at our facility with symptoms including vomiting, the passage of red currant jelly stool, and abdominal distension for 4 days. She subsequently experienced a seizure and underwent a right hemicolectomy for a perforated gangrenous ileocolic intussusception. Eighteen days later, the old twin presented with vomiting and the passage of red currant jelly stool for 1 day. She successfully underwent an ultrasound-guided hydrostatic reduction of her ileocolic intussusception.

***Conclusion*:** The exact cause of sequential twin intussusception is not entirely understood; however, it appears to be multifactorial. It seems that twins with similar systemic inflammatory responses, combined with congenital anatomical and genetic predispositions, are more likely to develop intussusception when simultaneously exposed to the same trigger.

Keywords: *sequential intussusception*; *twins*; *ultrasound hydrostatic reduction*

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Intussusception is the leading cause of intestinal obstruction in children in sub-Saharan Africa [1]. Intussusception is the telescoping or invagination of a proximal segment of the intestine and associated mesentery into an adjacent distal segment. The segment that invaginates is known as the intussusceptum, while the segment that receives it is called the intussuscipiens [1–4]. In 1674, Paul Barbette from Amsterdam described the first case of intussusception. Later, in 1793, Scottish surgeon James Hunter coined the term 'intussusception' [4, 5].

The incidence of intussusception ranges from 1 to 4 cases per 2,000 children worldwide [1–3]. Most reports indicate that it is more common in males. The peak incidence typically occurs between the ages of 4 and 9 months, which coincides with the weaning period from breast milk. Additionally, there are seasonal variations, with peaks often occurring during periods of acute viral infections [6].

Intussusception in children is primarily idiopathic, meaning the exact cause is often unknown. However, it can sometimes be associated with pathological lead points. A potential link exists between intussusception and respiratory or gastrointestinal viral infections. While intussusception is not regarded as a genetic disease, rare cases of familial occurrence have been reported, but these instances are considered incidental [2, 6–9]. There are various types of intussusception, with the ileocolic type being the most common, followed by the ileoileocolic type (Fig. 1) [2, 6]. The classical clinical triad of intussusception consists of intermittent abdominal pain, the passage of red currant jelly stool, and a sausage-shaped abdominal mass [2, 5, 10, 11]. The management of intussusception has evolved significantly over the years, shifting from mainly surgical interventions to nonoperative reduction techniques such as pneumatic, hydrostatic, and contrast methods, which have greatly decreased morbidity and mortality in infants [1, 3, 6].

Case presentation

A 5-month-old female infant, the younger of a set of dizygotic twins, presented to the children's emergency ward at University College Hospital in Ibadan with complaints of vomiting and the passage of red currant jelly stools for 4 days. She also exhibited abdominal distension, refusal to feed, and a low-grade fever. Additionally, she developed generalised clonic-tonic seizures 1 day prior to her referral to our hospital after presenting to two different hospitals, where she was treated with outpatient care. She was

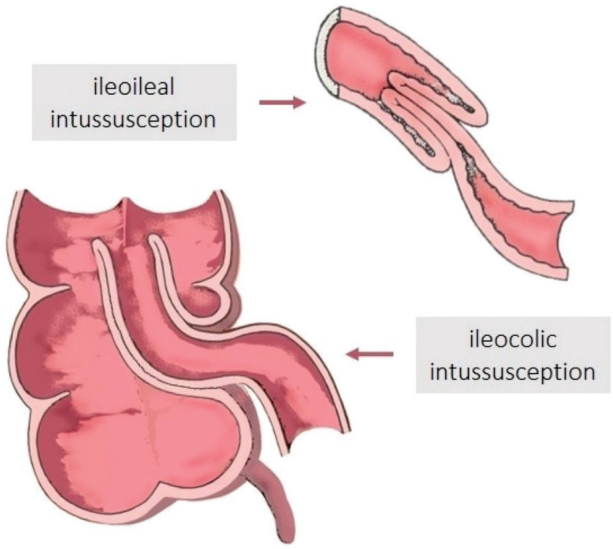

Fig. 1. Schematic drawing of ileocolic and ileoileal intussusceptions. Available at: https://epos.myesr.org/poster/esr/ecr2018/C2220/findings%20and%20procedure%20details.

delivered at term via spontaneous vaginal delivery, received age-appropriate immunisations, and was on mixed feeding. There is no known family history of intussusception.

Upon examination, the infant appeared dehydrated, febrile, and tachycardic. Her abdomen was distended, moved with respiration, and exhibited tenderness in the right upper quadrant. Bowel sounds were hyperactive, and a rectal examination revealed currant jelly stools.

An abdominal ultrasound scan showed multiple alternating concentric hypoechoic and hyperechoic rings (target sign) in the splenic flexure and descending colon in keeping with intussusception. There was also increased intraperitoneal fluid. A diagnosis of generalised peritonitis secondary to perforated ileocolic intussusception was made, after which the infant was resuscitated.

The patient underwent laparotomy, and intraoperative findings were ileocolic intussusception with a gangrenous apex at the ascending colon, as well as perforation on the antimesenteric border of the terminal ileum. No congenital abnormality was identified intraoperatively. Right hemicolectomy and ileocolic anastomosis were done. The postoperative period was uneventful, and she was discharged on postoperative day 6.

18 days later, the older of the set of dizygotic twins presented to the children's emergency ward at University College Hospital, Ibadan, with similar symptoms. She was brought in with complaints of vomiting and passage of red currant jelly stool for 1 day. The infant was delivered at term via spontaneous vaginal delivery, received age-appropriate immunisations and was on a mixed feeding regimen.

During the physical examination, she was found to have mild dehydration and a palpable mass in the right upper quadrant. Bowel sounds were hyperactive, and a rectal examination revealed red currant jelly stool.

A diagnosis of acute intestinal obstruction secondary to intussusception was made. An abdominal ultrasound confirmed the presence of ileocolic intussusception. The infant underwent ultrasound-guided hydrostatic reduction of the intussusception, which resolved her symptoms. A repeat abdominal ultrasound showed normal results, and she was allowed to begin oral intake. The patient was discharged on the second day of admission.

Discussion

Intussusception in children is a significant cause of morbidity and mortality, particularly in resource-poor countries. Timely diagnosis and treatment of intussusception significantly reduce the associated risks of morbidity and mortality [2]. The age of the twins in our case report is within the age range of previous reports [2, 6].

Delayed presentation remains a significant challenge in managing childhood intussusception in developing countries. This delay is often caused by misdiagnosis, as non-specialists can find it difficult to identify intussusception due to its nonspecific symptoms. This can lead to delayed referrals to specialists. Prompt referral to a paediatric surgeon may help reduce the need for surgery, as well as the associated morbidity and length of hospital stay [1, 6].

The management of intussusception has shifted from primarily surgical interventions to non-operative techniques, such as hydrostatic reduction and pneumatic reduction [3]. The second twin presented within a day of symptom onset, experienced no complications and had hydrostatic reduction. In contrast, the first twin had symptoms for 4 days and was complicated by multiple seizures, requiring surgical intervention and resulting in a longer hospital stay. This observation aligns with several studies that highlight delayed presentation as a significant challenge in managing childhood intussusception. The second twin was able to see a paediatric surgeon promptly due to the experience gained by the parents from the first twin's case, who had been taken to two different hospitals, where diagnosis was delayed before referral. This supports Serour's observation that parental experience with previous episodes of intussusception can reduce the delay between the onset of symptoms and definitive diagnosis, thereby facilitating more effective therapeutic measures and successful hydrostatic reduction [9].

The occurrence of intussusception in more than one patient within the same family is rarely reported [7]. The aetiology of idiopathic intussusception is considered incidental, and it has traditionally not been seen as having a hereditary basis. However, a genetic predisposition has been suggested in certain families [8]. In these families with a genetic predisposition, triggers such as viral infections or acquired immunity may make intussusception occur more easily. According to the literature, the risk of

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intussusception for siblings is estimated to be about 5 to 20 times higher than that of the general population [8].

Simultaneous intussusception in twins has been previously documented. Our literature search revealed no previously published reports of intussusception in twins from the sub-Saharan African continent. To our knowledge, this is the first case in sub-Saharan Africa. Thomas and Zachary reported a case involving a pair of identical twins, aged 2 years, who were diagnosed with intussusception 36 h apart. La Rosa F et al. reported a case of intussusception in a 3-month-old identical twin post vaccination with the Rotavirus vaccine [12]. Lee et al. also reported a case of simultaneous intussusception associated with adenovirus infection in a 1-year-old female identical twin [10]. Additionally, Nakanishi et al. reported that 18-month-old dizygotic twins simultaneously developed ileocecal intussusception following a generalised varicella infection [8].

Little is known about the aetiology of twin intussusception. Thomas and Zachary suggested that enlarged lymphoid tissue, often due to infection, is an important etiological factor. Nakanishi et al. proposed that anatomical enlargement itself might be the primary factor predisposing to intussusception. Meanwhile, Hsu et al. speculated that there could be a genetic predisposition to intussusception, potentially passed down in a pattern that favours its occurrence among siblings [8]. Mete et al. hypothesised that monozygotic twins may inherit similar responses to environmental factors due to anatomical predispositions to intussusception. Zhernakova et al. stated that genetic factors play a significant role in the development of immune-related disorders, as evidenced by the high concordance rates observed in monozygotic twin pairs and the increased familial clustering of these disorders. Bouma and Strober reported that an individual's genetic background influences their susceptibility to intestinal inflammation. Therefore, an inherited inflammatory response to the same infection in twins could lead to a similar degree of hypertrophy in the Peyer's patches [8].

Based on the literature and our experience, there is no single cause that explains the aetiology of idiopathic intussusception in twins. Instead, it seems to result from a combination of factors, including a similar systemic inflammatory response and the same congenital anatomic and genetic predispositions. It appears that the same environmental factors experienced simultaneously may lead to the onset of intussusception in twins [8].

Conclusion

Intussusception is a common cause of intestinal obstruction in children. The aetiology of simultaneous or concurrent twin intussusception is not fully understood, but it seems to be multifactorial. Delayed presentation remains a significant challenge in the management of childhood intussusception. Twins who share similar systemic inflammatory responses, as well as congenital anatomical and genetic predispositions, may simultaneously or concurrently develop intussusception when exposed to the same trigger at about the same time.

Conflict of interest

The authors declare no conflict of interest. The authors have not received any funding or benefits from industry or elsewhere to conduct this study.

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