Double-site antegrade and retrograde idiopathic intussusception in an infant: a case report and review of literature

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Intussusception is a very common surgical problem in infants. Double intussusception, however, is very rare in children. The authors report the successful management of a case of double-site antegrade and retrograde idiopathic intussusception in an 11-month-old boy. *Ann Pediatr Surg* 14:192–194 © 2018 Annals of Pediatric Surgery.

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Introduction

Intussusception is defined as the telescoping of a segment of bowel into an adjacent part. It is the most common cause of intestinal obstruction in infants [1]. About 90–95% of intussusceptions occur in children between the ages of 3 months and 3 years and are usually idiopathic with no pathological lead points [2]. Double intussusception is, however, a very rare occurrence [3,4]. We present a case of double-site antegrade and retrograde intussusception in an infant and reviewed the literature on the subject.

Case report

An 11-month-old boy presented with a 3-day history of excessive crying, persistent fever, and passage of mucus per rectum. At 3 h prior to presentation, he started passing bloody mucoid stools with bilious vomiting. Physical examination showed a sausage-shaped mass in the left lower abdominal quadrant and abdominal ultrasonography showed the target sign and did not detect the presence of more than one site of bowel invagination. Following a diagnosis of intussusception, the patient was resuscitated and planned for emergency laparotomy. Nonoperative reduction was not used in this case as the patient who presented late, 3 days after the onset of symptoms, was very ill, with persistent fever and a palpable mass in the left iliac fossa. This manner of presentation demonstrates the likelihood of bowel gangrene being present. At exploratory laparotomy, two sites of intussusception were found: a proximal, antegrade, ileocolic intussusception, and a distal, retrograde colocolic intussusception (Fig. 1). Both intussusceptions were separately reduced without any difficulty. There were no pathologic lead points. Postoperative recovery was uneventful.

Discussion

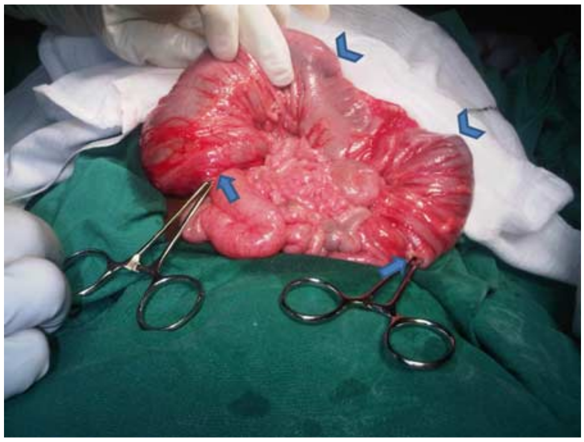
Intussusception is the most common cause of intestinal obstruction in infants older than 3 months. In this age group, it is most often ileocolic with no pathologic lead points [5]. Despite the frequent occurrence of idiopathic intussusception in children, double-site intussusception remains an extremely rare condition in this age group [3,4]. Shiu *et al.* [6] reported that double intussusception with pathological lead points was more common than idiopathic, double intussusception in children.

Current literature search showed 16 reported cases of double intussusception in the pediatric age group (Table 1). Eight were idiopathic, four had patent vitellointestinal ducts, and there was a case each of giant polypoid mass of heterotopic pancreas, submucous intestinal lipoma and hamartomatous polyps and a case of postoperative double intussusception following bilateral partial nephrectomy for Wilms tumor. Most cases of double intussusception reported in the literature occurred in adults with identifiable pathological lead points.

Chen *et al.* [4] described four subtypes of double intussusception, namely:

(1) Two separate intestines prolapsing into the same distal intestine, resulting in a characteristic triple

Fig. 1



Intraoperative photograph showing double intussusception: a proximal antegrade intussusception and a distal retrograde intussusception. Arrow: neck of intussusception. Chevron: apex of intussusception.

Table 1 Summary of reported cases of double intussusception

No.	References	Age	Sex	Site	Lead point	Treatment
1	Mustafa [7]	32 days	Male	Proximal and distal ileal loops	Patent vitellointestinal duct	Manual reduction
2	Him *et al.* [8]	7 months	Male	Ileocolic and ileocaecocolic (double compound intussusception)	Idiopathic	Manual reduction
3	Benson and Sparnon [9]	5 weeks	Male	Proximal and distal ileal loops	Patent vitellointestinal duct	Manual reduction
4	Scholz *et al.* [10]	11 years	Female	Double ileoileal	Giant polypoid mass of heterotopic pancreas	Manual reduction
5	Kiyan *et al.* [3]	8 months	Female	Ileocolic and colocolic	Idiopathic	Manual reduction
6	Kazez *et al.* [11]	8 years	Female	Colocolic and colocolic	Idiopathic	Manual reduction
7	Chen *et al.* [4]	4 years	Female	Jejunojejunal and ileocolic	Idiopathic	Manual reduction
8	Shiu *et al.* [6]	17 months	Male	Ileoileal and ileocolic	Idiopathic	Resection and anastomosis
9	Arnold *et al.* [5]	5 months	Female	Anterograde ileocolic and retrograde sigmoidocolic (double compound)	Idiopathic	Manual reduction
10	Destro *et al.* [12]	5 years	Male	Anterograde and retrograde ileoileal intussusception	Submucous intestinal lipoma	Reduction, resection and anastomosis
11	Wahid *et al.* [13]	11.5 months	Male	Jejunojejunal and ileoileal	Postbilateral partial nephrectomy	Manual reduction
12	Kim *et al.* [14]	20 days (preterm 23 weeks 1 day)	Female	Multiple small bowel intussusceptions	Idiopathic	Ileostomy and manual reduction
13	Mundada *et al.* [15]	23 days	Male	Proximal and distal ileal loops	Patent vitellointestinal duct	Resection and anastomosis
14	Davidson *et al.* [16]	15 years	Female	Mid-jejunum and distal ileum	Hamartomatous polyps	Small bowel resection and anastomosis following spontaneous reduction of intussusception
15	Park *et al.* [17]	80 days (preterm 25 weeks 6 days)	Male	Ileocolic and ileoileal	Idiopathic	Gastrograffin reduction and resection and anastomosis
16	Seid and Seman [18]	31 days	Male	Proximal and distal ileal loops	Patent vitellointestinal duct	Resection and anastomosis

circle sign on abdominal sonography and computed tomography scan [15].

(2) The double compound intussusception, which is extremely rare. Compound intussusception refers to double, treble, or quadruple intussusception occurring in a single mass [5].

(3) The double prolapse of the proximal and distal intestine through a patent vitellointestinal duct [7,9].

(4) Double-site intussusceptions as reported by Chen *et al.* [4] and in this report.

The exact mechanism for the formation of retrograde intussusception is unknown. Joseph and Desai [19] suggested that the weak antiperistaltic activity of the large bowel initiates the intussusception in a retrograde manner especially in the presence of an obstruction. Following initiation, the proximal bowel then slides over the intussuscepted area of the bowel through normal or exaggerated antegrade peristaltic waves. The predilection for the sigmoid colon is thought to favor the theory of initiation by the antiperistaltic waves in the left colon.

The treatment of choice for pediatric intussusception is nonoperative reduction. Failure to achieve reduction is often due to prolonged course with delayed presentation. In 5% of cases, it is because of a more complex anatomy requiring surgical reduction [5]. All reported cases of double intussusception required surgical intervention with good outcome.

Conclusion

Despite the rarity of double intussusception in children, the condition still occurs. There is a need for increased awareness of its existence by surgeons and sonologists as this will improve preintervention identification, aid in determining the best treatment modality, and eliminate delays from attempted nonoperative reduction.

Conflicts of interest

There are no conflicts of interest.

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