Simultaneous Antegrade and Retrograde Intussusceptions in a Child: A Rare Condition and Literature Review

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Abstract

Intussusception is one of the major abdominal emergencies in children under two years of age. Double intussusceptions are rare, and the simultaneous ante grade and retrograde occurrence is even rarer. Fewer than 10 such cases have been reported in the pediatric population.

An 8-month-old girl was presented to a peripheral health center with paroxysmal abdominal pain for two days, as well as postprandial vomiting and four mucous stools. She was diagnosed with amoebic dysentery. Faced with the appearance of glairy-bloody stools, she was admitted to our pediatric emergency unit. After examination and abdominal ultrasound, the diagnosis of intussusceptions was made. On the fourth day, operative findings showed a double ante grade and retrograde pattern. A manual reduction followed by an appendectomy was performed. The postoperative course was uneventful.

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Keywords

- Children
- Double intussusceptions
- Retrograde intussusceptions

Continuing education of peripheral practitioners will allow early diagnosis and appropriate treatment as this misdiagnosis could have compromised the vascular reserve of the bowel, resulting in intestinal ischemia and possibly perforation.

Introduction

Intussusception is defined as the telescoping of a segment of the bowel into an adjacent point. It is considered among the most common causes of acute abdominal pain in children less than two years of age.[1] Double intussusception is rare, and fewer than 20 cases have been reported in the English literature.[1,2] However, simultaneous antegrade and retrograde intussusception (SARI) is rarer in children, and most of these are reported in adult patients with a pathological lead point.[2,3] To our knowledge, this is the seventh case of SARI in children.[2,4-6] Here, we are reporting a first case of SARI in an Ivorian child because of the rarity of this condition. We will then discuss the difficulty of preoperative diagnosis and review the literature.

Case Presentation

An eight-month-old girl with no specific medical history was presented with paroxysmic abdominal pain for two days. She had six episodes of postprandial vomiting and four mucous stools with fever. The diagnosis of amoebic dysentery was made at the local hospital and the treatment consisted of metronidazole®, and paracetamol® and an oral dehydration solution. Faced with the appearance of glairy-bloody stools on the second day, her mother came to our emergency room. On admission, the infant was conscious and reactive. Her temperature was 38.2°C and she weighed 9kg. She was moderately dehydrated. The abdomen was breathing well, it was soft, compressible and painless. Abdominal palpation showed the mass in the umbilical quadrant. Digital rectal examination showed an empty rectum, and the finger of the glove was stained with blood. Cardiovascular and neurological examination was normal. The diagnosis of acute intestinal intussusception was suspected and confirmed by abdominal ultrasound **Figure 1**. Laboratory results show a hemoglobin level of 10.4g/dL and a prothrombin level of 97%. The blood ionogram was moderately disturbed. She was scheduled for emergency laparotomy. After the hydro-electrolyte rebalancing, a transverse laparotomy under the umbilical tissue was performed the next day. The operative findings revealed an antegrade ileocolic boudin through the ascending colon with multiple mesenteric adenopathies. During exploration, a second retrograde colocolic intussusception **Figure 2** was noted on the

transverse colon about 15 cm downstream of the first. Manual reduction was performed for both boudins, and the bowel appeared to be well perfused. There was no pathological lead point. An additional appendectomy was performed. The postoperative outcome was uneventful, and she was discharged home on the sixth postoperative day. Ten months later, there were no adverse events in the follow-up.

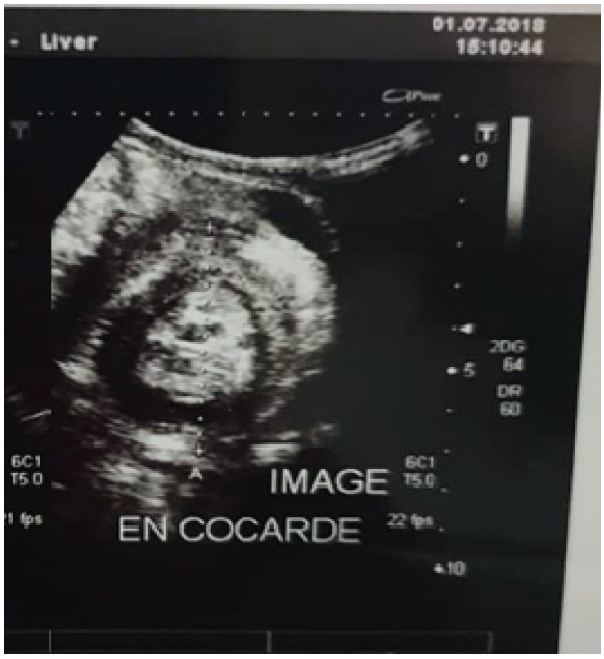

Figure 1. Abdominal ultrasonography show the classic target sign

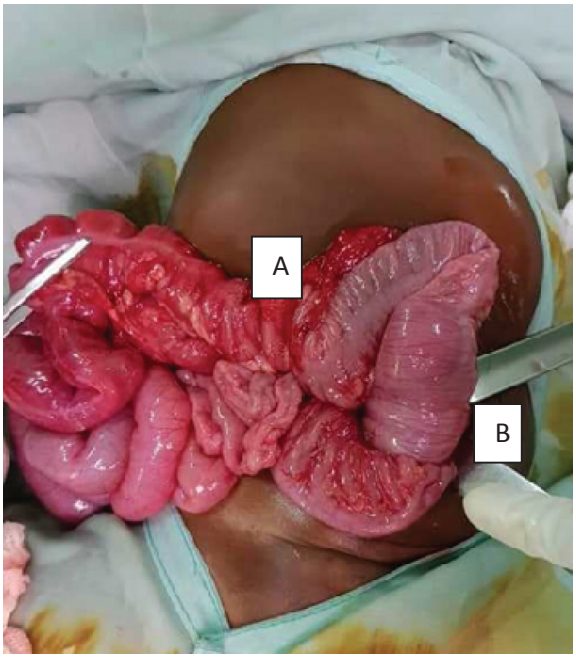

Figure 2. Operative findings shows double antegrade (A) and retrograde (B)

Discussion

Intussusception is one of the most common causes of bowel obstruction in children under 2 years of age, and antegrade ileocolic intussusception is the most common pattern.[5,7,8] Few cases of double intussusception have been reported. Among cases of double intussusception, SARI is a rare condition in children **Table 1**. A review of the literature over the past 30 years has identified fewer than 10 cases of SARI.[2,4-6,9] Our case was certainly the seventh case of SARI. This entity involving two distinct points in the bowel differs from compound intussusception which has been defined as double, triple or quadruple superimposed intussusception occurring as a single mass.[3-5,10] The term "retrograde intussusception" describes the intussusception of an intussusceptum in a proximal direction, but the exact mechanism of this condition remains unknown. Most retrograde forms are diagnosed in adults with a pathological lead point,[11] and the preferred site was the sigmoid colon. To date, apart from the known causes, the explanation of idiopathic retrograde forms is based only

on theories. The most common advanced theory was initiation by antiperistaltic waves in the left colon. In our case, the site of retrograde intussusception was the transverse colon which was free without attachment. We are unable to give an explanation for this condition.

Table 1. Reported cases of simultaneous antegrade and retrograde intussusception in children

Authors	Age / Gender	Type	Cause	Treatment
Shekhawat [7]	7 months ; M	Ileocolic and retrograde colocolic	Idiopathic	Manual reduction
Arnold [5]	5 months ; F	Double compound: ileocolic and retrograde sigmoido-colic	Idiopathic	Manual reduction
Lukong [6]	10 months ; M	Ileocolonic and retrograde colocolonic	idiopathic	Manual reduction
Egbuchulem [8]	11 months ; M	Double compound: ileocolic and retrograde colocolic	Idiopathic	Resection and anastomosis
Seyi-Olajide [3]	11 months ; M	Ileocolic and retrograde colocolic	Idiopathic	Manual reduction
Randimbinirina [9]	11 mois ; F	Ileocecocolic and retrograde sigmoidocolic	Idiopathic	Manual reduction
Our case	8 months ; F	Ileocolic and retrograde colocolic	Idiopathic	Manual reduction

Most authors have reported difficulties in the preoperative diagnosis of double intussusception. The clinical features in our case do not differ from those of classical intussusception (paroxysmal abdominal pain, palpable abdominal mass and redcurrant jelly stools). In our practice, amoebic dysentery is the most common diagnosis reported by most practitioners in peripheral health centers. This misdiagnosis could compromise the vascular supply of the bowel, resulting in intestinal ischemia and possibly perforation. In most cases of double intussusception such as ours, a single mass was palpated on abdominal examination, and ultrasound revealed the classic target sign. The second intussusception is not detected, either because there is too much gas distension or because the sonographer convinces himself of the diagnosis by finding the prominent intussusception and not looking for the others. None of the reported cases had two palpable masses on abdominal palpation, and less than half of them were diagnosed preoperatively by ultrasound or CT scan showing two target signs or a triple circle.[1,12-14]

To date, there is no medical treatment available for the preoperative diagnosis of double or retrograde invagination. The failure of the enema is undeniable. We

have systematically performed a laparotomy. On the other hand, laparoscopy was performed.[4] This approach limits parietal morbidity and the length of hospitalization. The operative results indicate a second intussusception with no pathological lead point. None of the previously reported SARIs had a pathological lead point.[1,6,9] Faced with the possibility of a double or compound or multiple intussusceptions, we agreed to always perform a careful evaluation of the entire bowel in order to identify the multiple distant segments that could be involved.

Conclusion

Simultaneous anterograde and retrograde intussusception is a rare entity in the pediatric population. Because of the potential for this occurrence, the radiologist should be referred by indicating this on the ultrasound bulletin. Coexistence of double or multiple intussusceptions requires careful examination of the entire bowel peroperatively.

Ethical Considerations

Written informed consent was obtained from the parents for publication of this case report and any accompanying images.

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Conflict of interests

There are no conflicts of interest.

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