

A CASE OF COMPOUND INTUSSUSCEPTIONS IN A NIGERIAN CHILD – A RARE FINDING IN A COMMON DISEASE

K.I. Egbuchulem¹, T.A. Lawal¹, M.C. Nweke² and A.O. Adeoye²

1. Division of Paediatric Surgery, Department of Surgery, University College Hospital Ibadan, Ibadan, Nigeria
2. Department of Pathology, University College Hospital, Ibadan, Nigeria

Correspondence:

Dr. T.A. Lawal

Department of Surgery,
University College Hospital,
PMB 5116, Ibadan 200212,
Nigeria
+2348069614811
Email: taiwo.lawal@hotmail.com

ABSTRACT

Intussusception is the most common, non-congenital, cause of intestinal obstruction in infants. There are various patterns of intussusception seen in children with the most common being ileo-colic variety. Compound intussusception in which intussusception involves more than one non-adjacent segment is a rare finding in the literature and it can be associated with worse morbidity than typically occurs, especially in a region where delayed presentation is a major contributor to morbidity and mortality in the treatment of intestinal obstruction.

We report the first documented case of double compound intussusception in an African child and reviewed relevant literature.

Keywords: Antegrade and retrograde; Children; Compound; Intussusception

INTRODUCTION

Intussusception is the invagination of a segment of intestine into the lumen of an immediately adjoining segment, which is mostly a distal segment with resultant intestinal obstruction. It is the most common acquired cause of intestinal obstruction in childhood¹ and can occur in variants including: ileo-ileal, colo-colic and ileo-colic, which are the most commonly seen type in 75 to 80%.^{2,3} Almost always, the invagination is from proximal to more distal segments of bowel and rarely, however, it may occur in a retrograde fashion. The rarest form of the pathology is compound intussusception in which intussusception involves both antegrade and retrograde patterns on different segments of bowel. Compound antegrade and retrograde intussusceptions are very rare and there had been no documentation in the English literature of such variety in an African child. This report is aimed at establishing the fact that while it is true that ileo-colic intussusception is the commonest form in children, a high index of suspicion should be developed by clinicians in looking out for rare forms as in our index case. We report the case of an 11-month-old Nigerian managed on account of double compound intussusception and reviewed relevant literature.

CASE PRESENTATION

An 11 month old boy was referred with a five day history of passage of watery stools, postprandial vomiting and a four day history of progressive abdominal distension associated with intermittent discomfort and inconsolable crying. There was

associated high grade fever and he passed red currant, jelly stools a few times before presentation. The patient had an episode of upper respiratory tract infection, which lasted a few days, two weeks prior to presentation.

Examination revealed an acutely ill child who was lethargic, febrile, pale and dehydrated. He also had tachycardia (140/minute) and tachypnoea (44/minute). The abdomen was distended with visible peristalsis but no generalized abdominal tenderness. A sausage shaped mass was palpated on the left hypochondrium with positive sign of Dance (absence of loops of bowels in the right half of the abdomen) and bowel sounds were hyperactive. There was no mass palpable in the rectum but the gloved finger was stained with mucoid blood stained faeces. A diagnosis of acute intestinal obstruction secondary to intussusception was made and the child was resuscitated.

Plain abdominal radiographs showed dilated loops of bowel with multiple air fluid levels. He was resuscitated with intravenous fluids, electrolyte, intravenous antibiotics and worked up for emergency exploratory laparotomy. The findings at surgery were: a retrograde colo-colic intussusception (proximal sigmoid colon into descending colon); antegrade ileo-colic intussusception as far distally as the proximal descending colon; perforations at the neck of both intussusceptions with devitalized bowel in between them and an area of necrotic bowel 10 cm from the neck of the ileo-colic

intussusception. There was minimal peritoneal soilage and about 50 millilitres of sero-fibrinous peritoneal fluid was drained. He had resection of both intussusceptions (Figure 1) and intervening devitalized bowel with an end to end primary ileo-sigmoid anastomosis.



Figure 1: Resected segment of ileum and colon

Postoperatively, he had oliguria, hypoproteinaemia and sepsis, but made good clinical progress and was discharged home.. He was followed up in the outpatient clinic over a three month period and had sustained clinical improvement.

Histopathology examinations of the resected proximal ileum and caecum showed multiple intussusceptions involving the sigmoid and descending colon (colo-

colic) and ileum up to the proximal descending colon (ileo-colic). There was marked distortion of the architecture of the appendix and intussusciens with extensive suppurative necrosis, vascular congestion and stromal oedema. Sections of the intussusceptions showed glandular epithelium overlying markedly hyperplastic lymphoid follicles with prominent germinal centres. Overall features were in keeping with intussusceptions with reactive follicular hyperplasia (Figure 2).

DISCUSSION

Intussusception or “introsusception” as it was called then, was first detailed in the literature in 1789 by John Hunter⁴ but the first surgery for a child with the condition was not reported until 1871 by Sir Jonathan Hutchinson.⁵ Intussusception, especially the idiopathic variety, is primarily a disease of childhood and is the commonest cause of intestinal obstruction between the ages of three months and two years.¹ While intussusception is quite common in childhood, double intussusception in which the invagination of bowel segment into adjacent ones occur at two segments of the intestine is rarely seen.⁶ Double intussusceptions can be one of four varieties. The first type is one in which two separate segments prolapse into the same distal loop of intestine, resulting in a characteristic “triple-circle” sign when sonography and computed tomography are performed.⁶ The second type, also called double compound intussusception⁷ involves both antegrade and retrograde intussusception in the same patient at the same time. This variety is extremely rare⁷ and mimics our index patient. The third type is

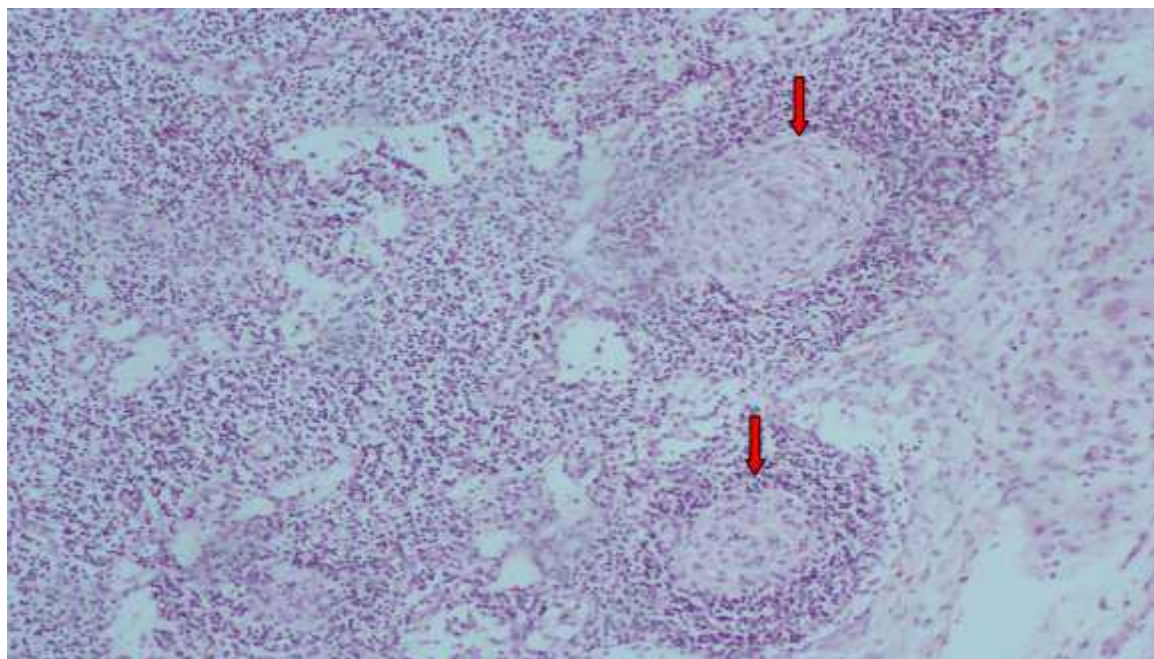


Figure 2: Section of the resected segment of the intussuscepted bowel showing a markedly hyperplastic lymphoid follicle with prominent germinal centres (arrow).

double prolapse of the proximal and distal intestine through a patent vitello-intestinal duct^{8,9} while the fourth type is antegrade intussusception involving two different sites on the bowel.¹⁰

In view of the rare nature of compound intussusception, there have been occasions when the second intussusception was found to have been missed at the time of surgery for the first intussusception even though both occurred synchronously.¹¹ This may be linked to the rarity of compound intussusception and the assumption in most instances that idiopathic intussusception of childhood is ileo-colic in location.¹¹ For intussusception to occur there must be bowel irritability and or a lead point. The aetiology of intussusception in early childhood remains a dilemma, as the majority of intussusception has no definite aetiological factor or demonstrable pathologic lead point^{10,12-14} unlike in older children and adults.¹⁵ We could not identify a pathologic lead point in our patient, rather the histological examination of the resected segment of bowel revealed markedly hyperplastic lymphoid follicles with prominent germinal centres suggestive of a secondary inflammatory response. Often, intussusception occurs during the time of weaning and may have a seasonal pattern that coincides with attacks of viral upper respiratory tract infection. The latter was noted in the presentation of the patient. The resected specimen also showed extensive lymphocytic infiltration, which is suggestive of a viral aetiology.

The major challenge in the treatment of intussusception in Africa is late presentation with associated complications such as dehydration, electrolyte imbalance, sepsis, bowel gangrene with peritonitis and shock¹⁶, which were present in our patient. In a related study on peritonitis in children, Osifo *et al* in Benin City, Nigeria, reported that management of childhood peritonitis in an African setting is challenging, because of late presentation, and intussusception accounted for 7.1% of all the causes of secondary bacterial peritonitis in that study.¹⁷

The management of intussusception in children in sub-Saharan Africa has not changed significantly and exploratory laparotomy is routinely performed following adequate resuscitation for such cases.¹⁸ Our patient who also had exploratory laparotomy and end to end bowel anastomosis was not an exemption. In a related study, comparing the rate of surgical intervention between colo-colic and ileo-colic intussusceptions in paediatric age group, it was observed that both groups have similar incidence of surgical intervention (82%) while resection rate was higher in the ileo-colic group compared to the colo-

colic group.¹⁹ In our patient, both intussusceptions were resected, because there was clinical evidence of gangrene and perforation, and an ileo-sigmoid anastomosis done.

CONCLUSION

Although, ileo-colic intussusception is the commonest pattern of intussusception in children, double compound intussusception is a rare disease of childhood that if not diagnosed early, may lead to extensive bowel loss. Intussusception is a paediatric surgical emergency hence, the onus lies on clinicians to look out for them and manage them effectively as this could affect the outcome.

This study was carried out in the Departments of Surgery and Histopathology, University College Hospital, Ibadan.

REFERENCES

1. **Ogundoyin OO**, Afolabi AO, Ogunlana DI, *et al*. Pattern and outcome of childhood intestinal obstruction at a Tertiary Hospital in Nigeria. *African Health Sciences* 2009; 9(3):170–173.
2. **Talabi AO**, Sowande OA, Etonyeaku CA and Adejuyigbe O. Childhood intussusceptions in Ile-Ife: What has changed? *Afr J Paediatr Surg* 2013; 10:239–242.
3. **Tandoh JFK**. Intussusception. In: Badoe EA, Archampong EQ, Da-Rocha-Afodu JT. Principles and practice of Surgery in the tropics. Ghana publishing corporation Accra: 2000. 3rd edition. Chapter 34, p.540–543.
4. **Hunter J**. On intussusception (read Aug 18, 1789). In: Palmer JF, ed. The Works of John Hunter, FRS London: Longman, Rees, Orme, Brown, Green, Longman, 1837:587–593.
5. **Hutchinson J**. A successful case of abdominal section for intussusception. *Proc R Med Chir Soc* 1873; 7:195–198.
6. **Kazez A**, Ozel SK, Kocakoc E and Kiris A. Double intussusceptions in a child: the triple circle sign. *J Ultrasound Med* 2004; 23:1659–1661.
7. **Him FP**, Weng YK and Hoi CW. A case of double compound intussusception in an infant. *Singapore Med J* 1980; 21:540–541.
8. **Benson JM**, Sparnon AL. Double intussusception of ileum through a patent vitello intestinal duct: report of a case and literature review. *Aust N Z J Surg* 1992; 62: 411–413.
9. **Mustafa R**. Double intussusception of the small bowel through a patent vitello-intestinal duct. *Br J Surg* 1976; 63:452.
10. **Kiyan G**, Tugtepe H, Iskit SH and Dagli TE. Double intussusception in an infant. *J Pediatr Surg* 2002; 37: 1643–1644.

11. **Yi-Hsin C**, Guan-Yeu D, Chih-Hau C, *et al.* Double site intussusception in a four-year-old girl. *J Med Science* 2006; 26(5):191–194.
12. **Kuremu RT**. Childhood intussusceptions at the Moi Teaching and Referral Hospital, Eldoret: Management challenges in a rural setting. *E Afr Med J* 2004; 81: 443–446.
13. **Carneiro PMR** and Kususi DM. Intussusception in children seen at Muhimbili National Hospital, Dar Es Salaam. *E Afr Med J* 2004; 81: 439–442.
14. **Edino ST**, Ochicha O, Mohammed AZ and Anumah M. Intussusception in Kano: a 5 year analysis of pattern, morbidity and mortality. *Niger J Med* 2003; 12: 221–224.
15. **Agha FP**. Intussusception in adult. *AJR Am J Roentgenol* 1986; 146 (3): 527–531.
16. **Gudeta B**. Intussusception in children: A ten year review. *E Afr Med J* 1993; 70:730–731.
17. **Osifo OD**, Ogiemwonyi SO. Peritonitis in children: Our experience in Benin City, Nigeria. *Surg Infect (Larchmt)* 2011; 12 (2): 127–130.
18. **Carneiro PMR** Editorial. Intussusception in the paediatric age group. *E Afr Med J* 2004; 81: 437–438.
19. **Grant HW**, Baccimazza I, Hadley GP. A comparison of colo-colic and ileo-colic intussusception. *J Pediatr Surg* 1996; 31(12):1607–1610.