Case Report

Journal of INTERNATIONAL MEDICAL RESEARCH

Journal of International Medical Research
2018, Vol. 46(8) 3427–3434

DOI: 10.1177/0300060518777337
journals.sagepub.com/home/imr
SAGE

Double simultaneous intussusception caused by Meckel's diverticulum and intestinal duplication in a child

Mingming Yu[#], Zhihong Fang[#], Jun Shen, Xiaomin Zhu, Dafeng Wang and Yingzuo Shi[#]

Abstract

Intussusception is common in children. Double simultaneous intussusception is a peculiar variety of intussusception with only 14 previously reported cases. We report a unique case of a child who suffered from double simultaneous intussusception with two lead points (Meckel's diverticulum and intestinal duplication). The patient was successfully treated with manual reduction along with resection of Meckel's diverticulum and intestinal duplication. The child recovered well.

Keywords

Double simultaneous intussusception, child, Meckel's diverticulum, intestinal duplication, laparotomy, abdominal pain

Date received: 1 February 2018; accepted: 23 April 2018

Introduction

Intussusception is a common aetiology of acute abdominal pain in children. Intussusception occurs in children of all ages, with predominance in children aged younger than 2 years old.[1–3] The most common intussusception is ileocolic intussusception, which accounts for approximately 98%. Double intussusception is rare. To date, only 14 cases of double intussusception (Table 1) have been reported.[4–17]

Department of Pediatric Surgery, Wuxi Children's Hospital, Wuxi, Jiangsu, China

[#]These authors contributed equally to this work.

Corresponding author:
Dafeng Wang, Department of Pediatric Surgery, Wuxi Children's Hospital, No. 229 Qingyang Rd, Wuxi, Jiangsu 210029, China.
Email: 79313136@qq.com

Table 1. Clinical data in children with double simultaneous intussusception.

Author	Age	Sex	Symptoms	Type	Treatment	Prognosis	Cause
Mustafa R, 1976[7]	32 days	Male	Bowel protruding through the umbilicus for 2 hours	Double ileo-vitelline duct	Manual reduction and resection of the vitellointestinal duct	Uneventful	Patent vitellointestinal duct
Him FP, 1980[8]	7 months	Male	Diarrhoea for 3 days and vomiting for 2 days	Double compound: ileocaecal and colocolic; ileocolic and colocolic	Manual reduction	Uneventful	Idiopathic
Bensen JM, 1992[9]	5 weeks	Male	Sudden prolapse of the bowel through the umbilicus	Double ileo-vitelline duct	Manual reduction and resection of the vitellointestinal duct	Uneventful	Patent vitellointestinal duct
Scholz S, 2000[10]	11 years	Female	Regular abdominal pain	Double ileoileal	Manual reduction and resection of the mass	Asymptomatic	Heterotopic pregnancy
Kıyan G, 2002[11]	8 month	Female	A 48-hour history of irritability, abdominal pain and current jelly-type rectal bleeding and bilious vomiting	Ileocolic and colocolic	Manual reduction	Uneventful	Idiopathic
Ahmet, K, 2004[12]	8 years	Female	Abdominal pain, vomiting, and bloody stool for 7 days	Double colocolic	Manual reduction	Uneventful	Idiopathic
Chen YH, 2006[4]	4 years	Female	Abdominal pain and bilious drainage after the first laparotomy	Jejunojejunal and ileocecal	First laparotomy: manual reduction of ileocecal intussusception; second laparotomy: resection of the jejunojejunal intussusception	Uneventful	Angiolipoma; idiopathic

(continued)

Table 1. Continued

Author	Age	Sex	Symptoms	Type	Treatment	Prognosis	Cause
Singh JK, 2009[13]	10 days	Male	Poor feeding, lethargy, and abdominal distension for 2 days	Ileoileal and ileocolic	Surgical resection	Died of coagulopathy and septicaemia on the 5th postoperative day	Idiopathic
Pandey A, 2010[14]	10 years	Male	Pain in the abdomen and no passage of flatus and faeces for 8 days, vomiting for 3 days	Jejunojejunal and ileoileal	Resection of jejunojejunal intussusception, manual reduction of ileoileal intussusception	Uneventful	Ileal polyp
Shiu JR, 2010[15]	17 months	Male	Painless haematochezia and anaemia for 1 day	Ileocolic and ileoileal	Manual reduction of the ileoileal intussusception; resection of the ileocolic intussusception	Uneventful	Idiopathic
Destro F, 2014[6]	5 years	Male	Seven-day history of cramping abdominal pain	Double ileoileal	Laparoscopy: manual reduction, resection of the tract containing the mass, and appendectomy	Uneventful	Lipoma
Wahid FN, 2014[16]	11.5 months	Male	Cramping abdominal pain and bilious vomiting 5 days after bilateral partial nephrectomies	Jejunojejunal and ileoileal	Manual reduction	Uneventful	Postoperative
Davidson J, 2015[17]	15 years	Female	Five-day history of intermittent and cramping abdominal pain with bilious	Not mentioned	Intussusceptions were resolved before laparotomy	Follow-up	Peutz–Jeghers syndrome

(continued)

Table 1. Continued

Author	Age	Sex	Symptoms	Type	Treatment	Prognosis	Cause
			vomiting and constipation				
Jolley H, 2017[5]	8 months	Female	Increasing abdominal distension, bilious emesis, and a bloody bowel on postoperative day 1	Double ileoileal	Manual reduction	Uneventful	Postoperative
Our case	21 months	Female	Paroxysmal abdominal pain for 1 day	Double ileoileal	Manual reduction, resection of Meckel's diverticulum, and intestinal duplication	Uneventful	Meckel's diverticulum and intestinal duplication

Occurrence of double simultaneous intussusception with two explicit leading points has not been previously reported.

In this report, we present a novel case of double simultaneous intussusception with two lead points (Meckel's diverticulum and intestinal duplication). We also review the epidemiology, pathogenesis, diagnosis, and therapies of this rare condition by analysing all previously reported cases.

Case presentation

A 21-month- old girl was referred to our hospital (Wuxi Children's Hospital, Wuxi, China) with a history of paroxysmal abdominal pain for 1 day. The parents of the patient denied any fever, vomiting, abdominal distension, diarrhoea, bloody urine, and faeces. Abdominal ultrasound showed that there were two concentric circles in the left and right lower abdomen (Figure 1). This finding indicated that there might be a double simultaneous intussusception. The result of abdominal computed tomography (Figure 2) was consistent with transabdominal ultrasound.

The patient initially received an air enema reduction, but this failed. The patient then received an exploratory laparotomy with emergency general anaesthesia. In the laparotomy, double ileoileal intussusception was found. One intussusception was approximately 65 cm from the ileocecal valve. The lead point of one of the intussusceptions was Meckel's diverticulum, which was approximately 5 cm in length and 1 cm in width, and it was connected with the normal mesentery. Meckel's diverticulum settled into the intestine approximately 15 cm along the basal part of the ileal wall. Another intussusception had intestinal duplication as the lead point and was approximately 110 cm from the cecum. The intestinal duplication was approximately 10 cm in length with a 2-cm-wide basal area and was interconnected

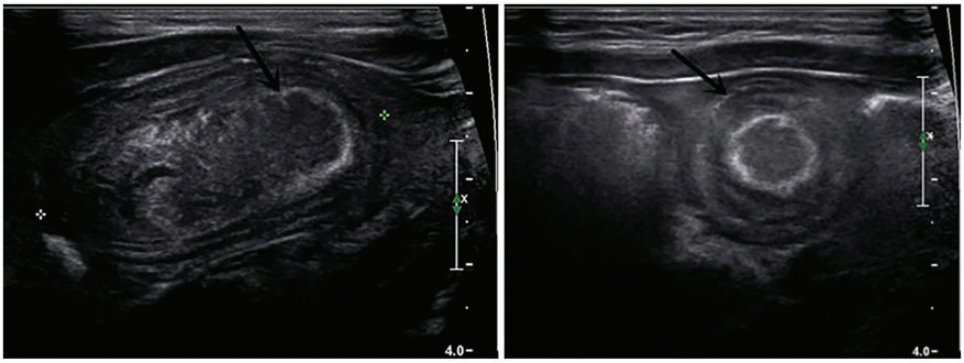

Figure 1. Abdominal ultrasound shows two concentric circles in the left and right lower abdomen (black arrow).

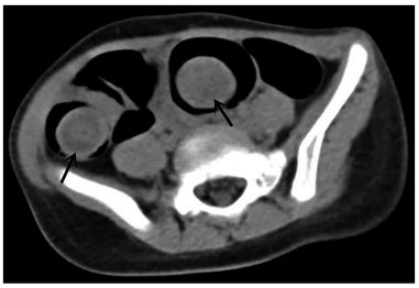

Figure 2. Abdominal computed tomography shows two intussusceptions in the left and right lower abdomen.

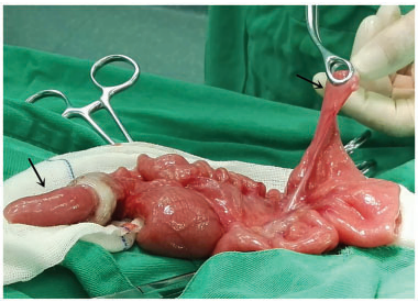

Figure 3. Photograph of surgery. The left black arrow shows Meckel's diverticulum, which is approximately 5 cm in length and 1 cm in width, and it is interconnected with the ileum without apparent mesenteric vessels. The right arrow shows intestinal duplication, which is approximately 10 cm in length with a 2-cm-wide basal area, and is connected with the normal mesentery.

with the ileum without apparent mesenteric vessels (Figure 3). There was partial necrosis in the intestinal intussusception. There were no other obvious abnormalities from the small intestine to the ligament of Treitz by further exploration. The two intussusceptions were successfully cured by manual reduction.

The patient then underwent resection of Meckel's diverticulum and intestinal duplication, as well as appendectomy. Postoperative pathology showed that Meckel's diverticulum had congestion and oedema with infiltration of lymphocytes and eosinophilic cells (Figure 4). The intestinal duplication was infiltrated by lymphocytes with a blind end, which contained heterotopic pancreatic tissue (Figure 5). Postoperative recovery of the patient was good, and there were no adverse events after a 6-month follow-up.

This article is a case report and literature review, and thus no ethical approval was required. We obtained written informed consent from the patient.

Discussion

Although intussusception is common in children, double simultaneous intussusception

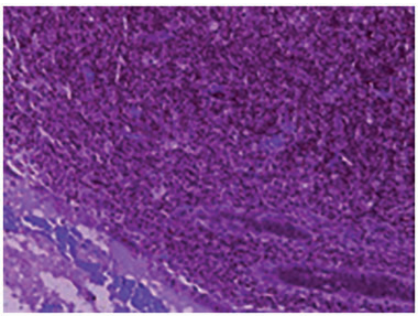

Figure 4. Postoperative pathology shows that Meckel's diverticulum has congestion and oedema with infiltration of lymphocytes and eosinophilic cells.

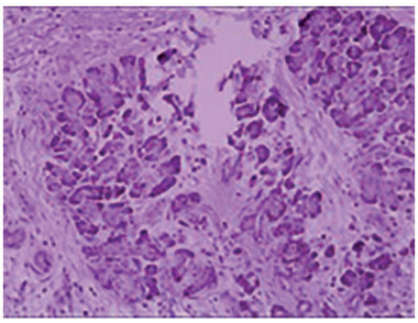

Figure 5. Postoperative pathology shows that the intestinal duplication is infiltrated with lymphocytes with a blind end, which contains heterotopic pancreatic tissue.

is a rare condition, which may be categorized into four types [4] These types include the following: two separate intestines prolapsing into the same distal intestine, resulting in a characteristic "triple-circle" sign by sonography; two separate intussusceptions in two separate sites; double compound intussusception; and double prolapse of the proximal and distal intestine through a patent vitellointestinal duct. Only 14 cases of double intussusception have been reported.[4–17]

Double simultaneous intussusception may be mainly attributed to an explicit lead point, the idiopathic status, or even the postoperative status. The lead point is the main factor, including a patent vitellointestinal duct, heterotopic pregnancy, ileal polyp, angiolipoma, lipoma, and Peutz–Jeghers syndrome. Most of the previously reported cases showed only one lead point. However, this is the first case of double intussusception with two lead points. Additionally, five patients with double intussusception were idiopathic. The postoperative status is another factor that should be taken into consideration. Two previous cases of double intussusception occurred after initial operations within 5 days, including exploratory[5] laparotomy and bilateral partial nephrectomy.

The clinical features of double simultaneous intussusception mainly consist of abdominal pain, abdominal distension, vomiting, and bloody stool, similar to common intussusception to some extent.[18,19] However, some other concomitant symptoms should not be ignored, such as diarrhoea, poor feeding, irritability, and lethargy, especially in infants. The proximal and distal intestines passing through a patent vitellointestinal duct is a special type of prolapse of double intussusception, which is rare with unambiguous features.

The diagnosis of double intussusception mainly depends on the medical history, a physical check-up, and an auxiliary examination. The medical history refers to clinical manifestations discussed above. The most useful sign of a physical check-up is an abdominal mass. An abdominal mass was apparent in less than half of the reported cases of double intussusception[6,8,10,11,13,16] and none of them presented as two palpable masses. Some of these patients can have abdominal tension and increased bowel sounds. Abdominal ultrasound is the most common in auxiliary examination for double intussusception and the main feature is double concentric circular or triple-circle sign, which is also apparent in less than half of the cases. Additionally, X-ray

and computed tomography are important auxiliary methods.[20] Among all of the cases of double intussusception, only eight patients (including our patient)[5,7,9,12,14,15,17] were diagnosed before surgery, while the remaining seven patients were diagnosed during intraoperative inspection.

Treatment of double simultaneous intussusception is different from common intussusception to some extent. Most intussusceptions can be treated with non-surgical methods, such as enema with barium, air, or water. In all of the 15 cases of double simultaneous intussusception, most children accepted enema with air or water, but all of these treatments failed. This failure could have been due to the structure of double simultaneous intussusception, which is more complex and tighter than just intussusception. Therefore, we suggest that children who are diagnosed with double simultaneous intussusception should accept surgery directly. This could avoid unnecessary treatment, as well as wasting time and decreasing the risk of bowel necrosis. Surgical methods include laparotomy (majority) and laparoscopic surgery.[21,6]

Postoperative recovery of most children with double simultaneous intussusception is usually good. Only one newborn with double simultaneous intussusception died because of coagulopathy and septicaemia.[13] Therefore, clinical doctors need to pay more attention to patients after surgery, especially to younger patients with a poor physical condition. Follow-up after discharge from hospital after treating double simultaneous intussusception is also important.

Conclusion

In conclusion, double simultaneous intussusception is rare. To the best of our knowledge, this is the first case of double simultaneous intussusception caused by Meckel's diverticulum and intestinal duplication in a child. The diagnosis of this condition mainly depends on the medical history, a physical check-up, and an auxiliary examination, while only approximately half of these cases can be diagnosed before surgery. Generally, non-surgical methods are invalid for double simultaneous intussusception and emergent exploration is suggested. Most patients can recover well after surgery and postoperative follow-up is recommended. With further development of technology, more similar cases of double simultaneous intussusception are likely to be reported.

Declaration of conflicting interests

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The authors disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: This research was supported by Wuxi Young Medical Talents (Grant No. QNRC023) and the Medical Innovation Team of Jiangsu Province (Grant No. CXTDB2017016).

References

1. Kawasaki Y, Shinozaki S, Yano T, et al. Intussusception due to an inverted Meckel's diverticulum diagnosed by double-balloon enteroscopy. *Case Rep Gastroenterol* 2017; 11: 632–636. 2017/12/29. DOI: 10.1159/000481161.
2. Jiang J, Jiang B, Parashar U, et al. Childhood intussusception: a literature review. *PloS One* 2013; 8: e68482. 2013/07/31. DOI: 10.1371/journal.pone.0068482.
3. Edwards EA, Pigg N, Courtier J, et al. Intussusception: past, present and future. *Pediatr Radiol* 2017; 47: 1101–1108. 2017/08/06. DOI: 10.1007/s00247-017-3878-x.

4. Chen YH, Diau GY, Chang CH, et al. Double site intussusception in a four-year-old girl. *J Med Sci* 2006; 26: 191–193.
5. Jolley H, Gefen AM, Ginsburg H, et al. Double ileoileal intussusception following surgical reduction of ileocolic intussusception in an 8-month-old female. *J Pediatr* 2017; 186: 208.
6. Francesca D, Noemi C, Michela M, et al. An interesting case of double compound intussusception without intestinal occlusion in a 5-year-old boy. *European J Pediatr Surg Rep* 2014; 2: 20–22.
7. Mustafa R. Double intussusception of the small bowel through a patent vitello-intestinal duct. *Br J Surg* 1976; 63: 452.
8. Him FP, Weng YK and Hoi CW. A case of double compound intussusception in an infant. *Singapore Med J* 1980; 21: 540–541.
9. Benson JM and 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.
10. Scholz S, Loff S and Wirth H. Double ileoileal intussusception caused by a giant polypoid mass of heterotopic pancreas in a child. *Eur J Pediatr* 2000; 159: 861–862.
11. Kiyan G, Tugtepe H, Iskit SH, et al. Double intussusception in an infant. *J Pediatr Surg* 2002; 37: 1643.
12. Kazez A, Ozel SK, Kocakoc E, et al. Double intussusception in a child. *J Ultrasound Med* 2004; 23: 1659–1661.
13. Singh JK, Bawa M, Kanojia RP, et al. Idiopathic simultaneous intussusceptions in a neonate. *Pediatr Surg Int* 2009; 25: 445–447.
14. Pandey A, Rawat JD, Wakhlu A, et al. Simultaneous occurrence of jejuno-jejunal and ileo-ileal intussusception in a child: a rare occurrence. *BMJ Case Rep* 2011; 2011: bcr0820103294.
15. Shiu JR, Chao HC, Chen CC, et al. Rare concurrent ileoileal and ileocolic intussusceptions in a child presenting with painless hematochezia. *Pediatr Neonatol* 2010; 51: 359–362.
16. Wahid FN, Malkan AD, Mccarville MB, et al. Double small bowel intussusception complicating bilateral partial nephrectomies. *J Pediatr Surg Case Rep* 2014; 2: 30–32.
17. Davidson J, Wright NJ and Kufeji D. Differential diagnosis of double site intussusception in childhood: a 15-year-old girl presenting with bowel obstruction. *BMJ Case Rep* 2015; 2015.
18. Yu ML, Lee KH and Li YL. The crescent-in-doughnut sign in intussusception. *Abdom Radiol (NY)* 2018; 43: 1266–1267.
19. Gluckman S, Karpelowsky J, Webster AC, et al. Management for intussusception in children. *Cochrane Database Syst Rev* 2017; 6: CD006476.
20. Di Grezia G, Gatta G, Rella R, et al. Abdominal hernias, giant colon diverticulum, GIST, intestinal pneumatosis, colon ischemia, cold intussusception, gallstone ileus, and foreign bodies: our experience and literature review of incidental gastrointestinal MDCT findings. *Biomed Res Int* 2017; 2017: 5716835. DOI: 10.1155/2017/5716835.
21. Kleizen KJ, Hunck A, Wijnen MH, et al. Neurological symptoms in children with intussusception. *Acta Paediatrica* 2014; 80: 293–298.