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

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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

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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

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| Author          | Age  | Sex    | Symptoms                                                                 | Type                          | Treatment                                      | Prognosis   | Cause                          |
|-----------------|------|--------|---------------------------------------------------------------------------|-------------------------------|-----------------------------------------------|-------------|--------------------------------|
| Mustafa R, 1976 | 7    | Male   | Bowel protruding through the umbilicus for 2 hours                        | Double ileo-vitellointestinal duct | Manual reduction and resection of the vitellointestinal duct | Uneventful  | Patent vitellointestinal duct  |
| Him FP, 1980    | 7    | 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 | 5    | Male   | Sudden prolapse of the bowel through the umbilicus                        | Double ileo-vitellointestinal duct | Manual reduction and resection of the vitellointestinal duct | Uneventful  | Patent vitellointestinal duct  |
| Scholz S, 2000  | 11   | Female | Regular abdominal pain                                                    | Double ileoileal               | Manual reduction and resection of the mass     | Asymptomatic | Heterotopic pregnancy          |
| Kyan G, 2002    | 8    | 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  | 8    | Female | Abdominal pain, vomiting, and bloody stool for 7 days                     | Double colocolic               | Manual reduction                               | Uneventful  | Idiopathic                     |
| Chen YH, 2006   | 4    | 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)
| 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 16 | 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)
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

| Author          | Age   | Sex | Symptoms                                      | Type                           | Treatment                                                                 | Prognosis | Cause                                      |
|-----------------|-------|-----|-----------------------------------------------|-------------------------------|---------------------------------------------------------------------------|-----------|--------------------------------------------|
| Jolley H, 2017  | 8 months | Female | Vomiting and constipation, abdominal distension, bilious emesis, and a bloody bowel | Double ileoileal intussusception | Manual reduction, resection of Meckel’s diverticulum and intestinal duplication | Uneventful | Meckel’s diverticulum and intestinal duplication |
| Our case        | 21 months | Female | Paroxysmal abdominal pain for 1 day           | Double ileoileal intussusception | Manual reduction, resection of Meckel’s diverticulum and intestinal duplication | Uneventful | Meckel’s diverticulum and intestinal duplication |

Table 1. Continued
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
is a rare condition, which may be categorized into four types. These types include: 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.

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 laparotomy and bilateral 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. 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 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. Among all of the cases of double intussusception, only eight patients (including our patient) 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.

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. 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.

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