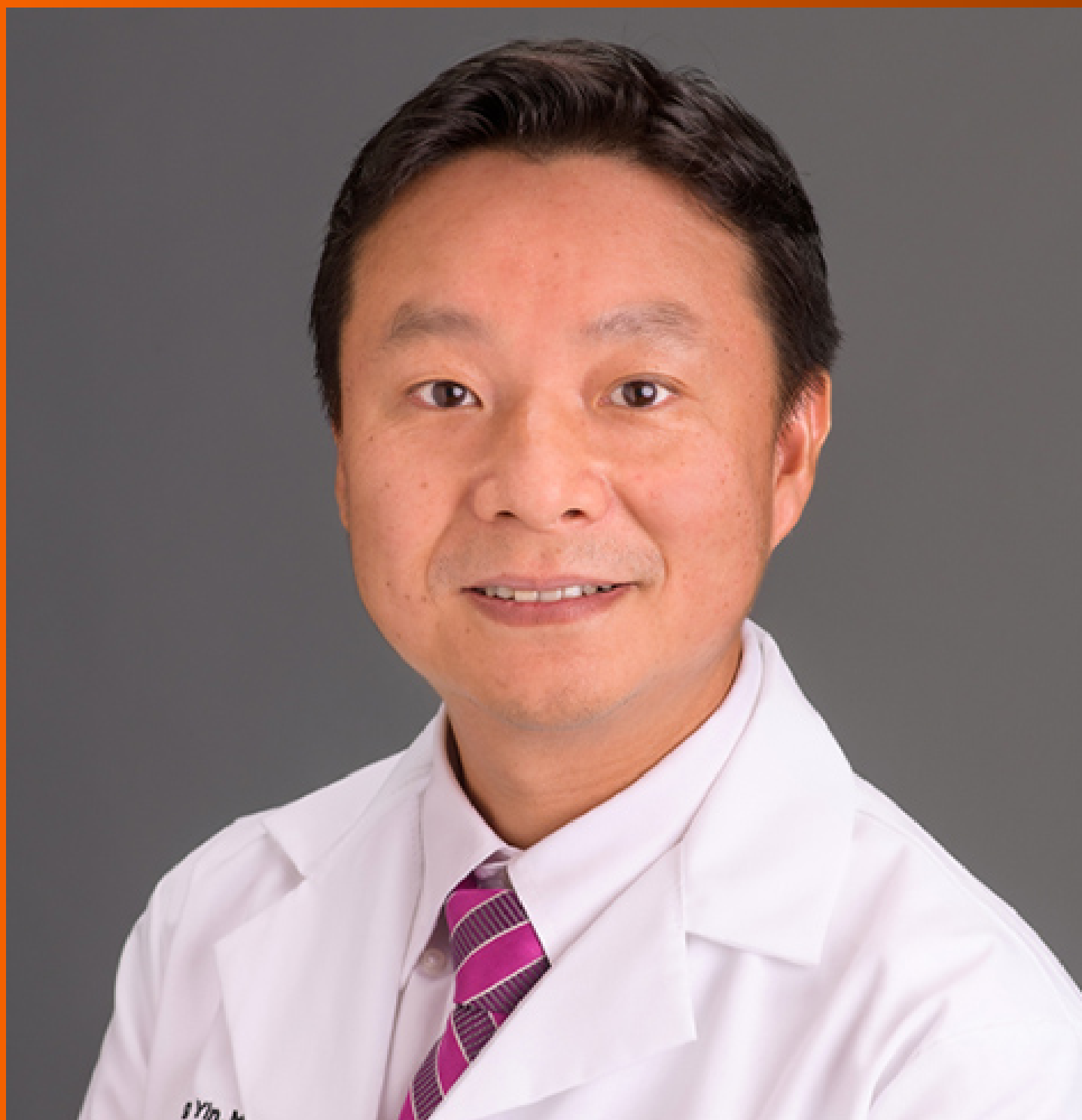


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Intestinal intussusception caused by intestinal duplication and ectopic pancreas: A case report and review of literature

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Abstract

BACKGROUND

Intestinal intussusception caused by intestinal duplication and ectopic pancreas is extremely rare in the clinic and has not been reported previously.

CASE SUMMARY

A 29-year-old man was admitted to the hospital for chronic abdominal pain and bloating. The preoperative diagnosis was intestinal obstruction and intussusception. Then, laparotomy, partial small intestinal resection and extraintestinal decompression were performed. Postoperative pathology confirmed intestinal duplication and ectopic pancreas. After surgery, the patient recovered well with no complications. No recurrence was observed after more than 5 mo of follow-up.

CONCLUSION

We report a new case of a young male with intussusception caused by intestinal duplication and ectopic pancreas. Surgery is the main treatment for these conditions. This study aimed to raise awareness and provide information to improve the clinical management of this rare yet serious condition.

Key Words: Intestinal intussusception; Ectopic pancreas; Intestinal duplication; Intestinal obstruction; Adult; Case report

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Core Tip: We present a new case of a young man with intussusception caused by intestinal duplication and ectopic pancreas. This case highlights that intestinal duplication and heterotopic pancreas should be considered causes of intestinal obstruction associated with intussusception.

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INTRODUCTION

Intestinal intussusception refers to intestinal tube insertion into the connected intestinal lumen, which causes obstruction of the intestinal contents. Adult intussusception only accounts for 5% of the total incidence[1]. The average age of adult patients is 50 years, and the male-to-female ratio is 1:5[2]. Adult intussusception has no specific clinical manifestations and lacks the classic triad of pediatric intussusception symptoms: abdominal pain, vomiting, and jam-like stool. Abdominal computed tomography (CT) is considered the most useful examination, but most specific causes still require intraoperative detection or postoperative pathology for clarification. Ninety percent of adult intussusception is caused by tumors, polyps, diverticulum, *etc.*[3-6]. Thus, 70% to 90% of adult intussusception requires surgical treatment[7]. However, to date, there are no case reports on intussusception caused by small intestinal duplication and ectopic pancreas.

We report a new case of a young man with intussusception caused by intestinal duplication and ectopic pancreas. Informed consent for the publication of these data was obtained from the patient.

CASE PRESENTATION

Chief complaints

A 29-year-old man was admitted to the hospital for repeated abdominal pain and bloating for more than 2 mo that had been aggravated for 2 d.

History of present illness

The patient had repeated abdominal pain and abdominal distension for 2 mo and had been treated in many hospitals. The symptoms could be relieved by symptomatic treatments, such as rehydration and pain relief. Due to aggravation of the above symptoms for 2 d, the patient was admitted to our hospital to consider the possibility of intestinal obstruction. Since onset, the patient's anal exhaust and defecation had decreased, with no additional vomiting, chills, fever or other discomfort. A yellow-brown stool was discharged once on the day of admission, and no mucous or blood was present in the stool.

History of past illness

The patient had no relevant previous medical history.

Personal and family history

The patient's family history was unremarkable.

Physical examination

The physical examination findings were as follows: slight abdominal bulge, no obvious tension of the abdominal muscles, right lower abdominal tenderness, slight rebound pain, negative Murphy sign, and no obvious palpable abdominal mass.

Laboratory examinations

Initial laboratory data revealed a white blood cell count of $8.3 \times 10^9/L$, neutrophil count of $6.67 \times 10^9/L$, neutrophil percentage of 83.1% and C-reactive protein of 13.9 mg/L. Liver and kidney parameters were normal.

The postoperative pathology results were as follows: Two intestinal canal-like structures were observed in the mesentery-side lumen, measuring approximately 6 cm in length and 2.5 cm in diameter and 2.5 cm in length and 2 cm in diameter. The intestinal canals were observed directly after incision (Figure 1). Microscopically, the intestinal mucosa, submucosa, muscular layer and serosal layer were observed in both intestinal canals. The muscular layer of the short intestinal wall was significantly thickened, and local dysplasia of the external longitudinal muscle was accompanied by disordered

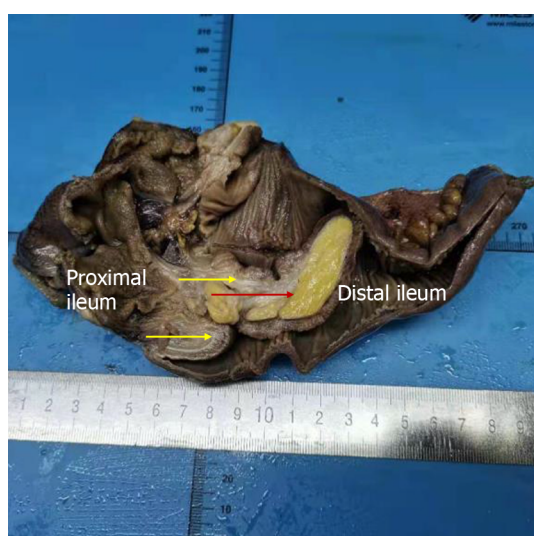


Figure 1 Postoperative gross specimen showing intestinal duplication (yellow arrow) and adipose tissue hyperplasia with ectopic pancreatic tissue (red arrow).

growth of the muscular bundle (Figure 2A). The serosal surface of the long intestinal canal showed fatty tissue hyperplasia with ectopic pancreas, and pancreatic acinar and islet tissue and pancreatic duct cells were observed (Figure 2B and C).

Imaging examinations

Abdominal enhanced CT showed an annular bowel shadow in the lower left abdomen, indicating the possibility of intussusception, intestinal wall thickening, and inflammatory edema (Figure 3).

FINAL DIAGNOSIS

The following diagnoses were made: Intestinal obstruction, intestinal duplication and ectopic pancreas, and intussusception.

TREATMENT

The patient underwent emergency exploratory laparotomy, partial small intestinal resection and extraintestinal decompression.

During the operation, the proximal small intestine was found to be inserted into the distal small intestine on the ileum approximately 80 cm away from the ileocecal valve. The lesion length was approximately 20 cm, and the proximal small intestine was dilated to a diameter of approximately 8 cm. The distal small intestine was empty, and the small intestine at the site of intussusception was edematous. Some of the intestinal wall was thickened and hardened, resulting in complete obstruction of the intestinal cavity. Exploration and manual reduction of a mass measuring approximately 3 cm × 2.5 cm at the site of intussusception were difficult. Small intestinal tumors, intestinal intussusception and intestinal obstruction were considered. Partial small intestinal resection and extraintestinal decompression were performed.

OUTCOME AND FOLLOW-UP

The patient recovered well with no complications. No recurrence was found after more than 5 mo of follow-up.

DISCUSSION

Intussusception is a special form of intestinal obstruction, accounting for approximately 1%-5% of cases [1]. The exact cause of approximately 8%-12% of adult intussusception is unclear [1,7,8]. Any intestinal

Table 1 Adult intestinal intussusception and ectopic pancreas

Ref.	Gender	Age	Location
Ganapathi <i>et al</i> [22], 2011	Male	26	Ileo-ileal
Sciannamea <i>et al</i> [23], 2020	Female	33	Ileo-ileal
Gold <i>et al</i> [24], 2020	Female	23	Intestinal
Giordano <i>et al</i> [25], 2017	Male	29	Jejunojejunal
Chuang <i>et al</i> [26], 2010	Female	26	Ileo-ileal
Abe <i>et al</i> [27], 2020	Female	43	Ileo-ileal
Daniel <i>et al</i> [28], 2021	Female	78	Jejunojejunal

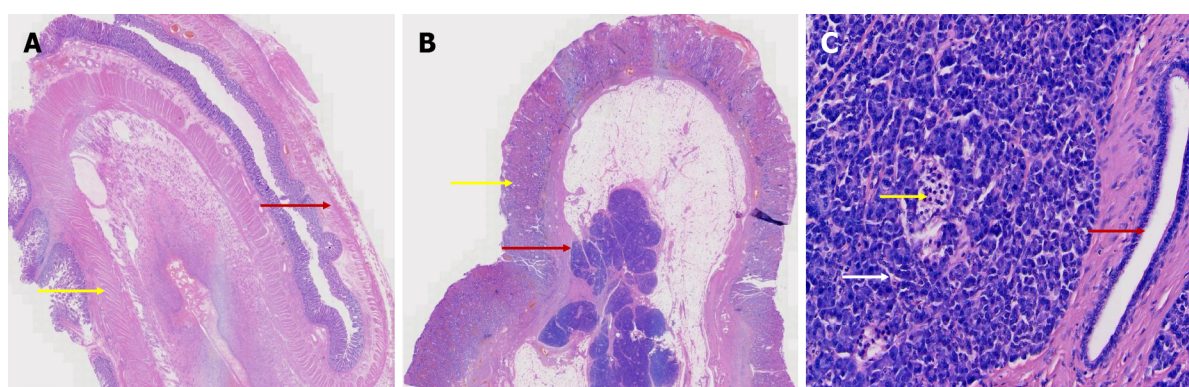


Figure 2 Hematoxylin & eosin staining. A: Microscopic view of the wall of the normal intestine (red arrow) and duplicated intestine (yellow arrow) [hematoxylin & eosin (HE) \times 5]; B: Microscopic view of adipose tissue hyperplasia on the serosa surface of the duplicated intestinal wall (yellow arrow) with ectopic pancreas (red arrow) (HE \times 5); C: Microscopically, the ectopic pancreatic tissue showed pancreatic islet cells (yellow arrow), pancreatic acinus (white arrow), and the pancreatic duct (red arrow) (HE \times 200).

disease that changes the normal peristalsis of the intestine increases the risk of intussusception[9]. Adult intussusception is often secondary to intestinal tumors, polyps, diverticulum, *etc.* Therefore, for suspected adult intussusception patients, digestive tract radiography, CT, magnetic resonance imaging and other examinations should be actively performed to clarify the etiology, and active surgery is needed to treat the primary disease and relieve the obstruction. For patients with adult intussusception that can be reduced, subsequent resection of the organic lesions remains the preferred treatment.

Intestinal duplication and ectopic pancreas are two independent congenital defects. The incidence of ectopic pancreas according to autopsy reports is 0.5%-13%[10]. The etiology of ectopic pancreas is still unclear. Studies have shown that during embryonic development, pancreatic primordia adhere to or penetrate the intestinal wall of the embryo and continue to develop and form in various abnormal locations with the movement of intestinal transposition[11,12]. Ectopic pancreas is mainly composed of pancreatic duct and acinar tissue and usually lacks islet tissue. There are no anatomical or vascular connections between the ectopic pancreas and normal pancreas[13]. It most commonly occurs in the duodenum but can also appear in the jejunum and, rarely, in the ileum[14]. Ectopic pancreas is generally considered asymptomatic[15]. In a few case reports, intestinal obstruction, abdominal pain and bleeding were also mentioned[16,17]. Due to the diverse locations and lack of specific lesions, early ectopic pancreas diagnosis is difficult, easily leading to misdiagnoses or missed diagnosis. We searched the literature on PubMed for adult intussusception and ectopic pancreas, and the results are summarized in Table 1.

The incidence rate of intestinal duplication is 1 in 10000 newborns, and it is rare in adults[18]. We retrieved the literature on adult intussusception caused by intestinal duplication from PubMed, and the publications are listed in Table 2. The differences between intestinal duplication and Meckel's diverticulum are as follows: (1) Intestinal duplication shares a blood supply with the surrounding intestine, while diverticulum has an independent blood supply; and (2) well-developed smooth muscle is present in intestinal duplication but not in Meckel's diverticulum[19]. Studies suggest that there is a direct communication hole between intestinal duplication and the normal intestinal wall for unknown reasons[20,21]. With the closure of the hole, the muscular layer becomes discontinuous, and feces form a mucosal bridge after passing through the hole, which leads to the occurrence of adult intussusception.

Table 2 Adult intestinal intussusception caused by intestinal duplication

Ref.	Gender	Age	Location
Kimura <i>et al</i> [21], 2018	Male	19	Colon
Ho[29], 2012	Male	25	Terminal ileum, appendix, colon and rectum
Li <i>et al</i> [30], 2013	Male	25	Ileum
Al-Qahtani[31], 2016	Female	32	Ileum
Kyo <i>et al</i> [32], 2016	Male	20	Colon
Kim <i>et al</i> [33], 2014	Female	19	Ileal
Reiser-Erkan <i>et al</i> [34], 2010	Male	25	Colon
Kusnierz <i>et al</i> [35], 2014	Unknown	31	Duodenal
Nadatani <i>et al</i> [36], 2016	Male	73	Ileal
O'Connor <i>et al</i> [37], 1999	Female	32	Duodenal

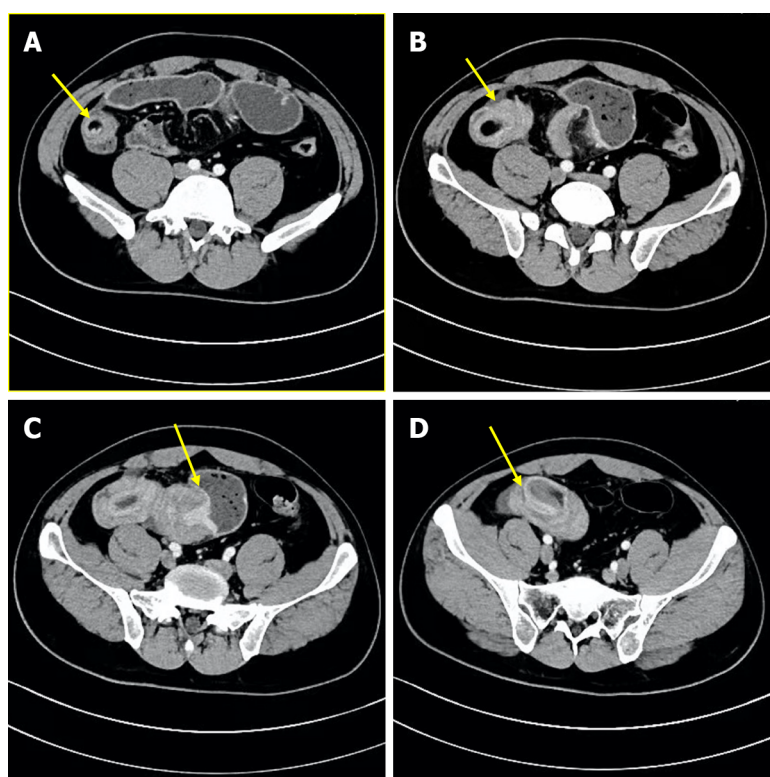


Figure 3 Abdominal enhanced computed tomography showing an annular bowel shadow in the lower left abdomen, indicating the possibility of intussusception and intestinal wall thickening. A: Head of intussusception; B: Distal sheath of intussusception; C: Neck of intussusception; D: Sheath of intussusception.

Although adult intussusception, ectopic pancreas and intestinal duplication have been reported separately, our case was unique, as intussusception caused by intestinal duplication and heterotopic pancreas has not been reported previously.

In this case, the patient had been treated in many hospitals for repeated abdominal pain and received symptomatic treatments, such as fluid infusion and pain relief. However, because the fundamental cause of the disease was not determined, the symptoms persisted, and intussusception inevitably occurred, leading to complete intestinal obstruction. Therefore, we speculate that due to the abnormal anatomical position, when intestinal duplication is combined with ectopic pancreatic tissue, the ectopic pancreatic tissue is accompanied by the continuous growth of adipose tissue. Because of the compensatory ability of the body, increased intestinal peristalsis occurs, resulting in intussusception and complete intestinal obstruction. Therefore, early correct diagnosis and timely treatment are critical for adult intussusception.

CONCLUSION

This is the first reported case of intestinal intussusception caused by intestinal duplication and ectopic pancreas. Therefore, intestinal duplication and ectopic pancreas should be considered in the differential diagnosis of intussusception. Although modern diagnostic technology has greatly progressed, diagnosis of intestinal duplication and ectopic pancreas remains difficult. Definitive diagnosis usually depends on postoperative pathology.

FOOTNOTES

Author contributions: Wang TL, Gong XS and Wang J contributed equally to this work; Wang TL collected the data of the case and drafted the manuscript; Gong XS and Wang J performed the surgery; Wang J assisted in the revision; and Long CL reviewed the manuscript and provided constructive input; all authors read and approved the final manuscript.

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