Journal of
Medical Case Reports

CASE REPORT

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A case of Meckel's diverticulum complicated with intestinal duplication: a case report

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Abstract

Background Meckel's diverticulum and intestinal duplication malformations are two isolated digestive tract malformations in children. It is uncommon to see cases of both digestive tract malformations occurring at the same time. This report presents a rare case of Meckel's diverticulum complicated with intestinal duplication, highlighting the importance of intraoperative exploration.

Case presentation A 5-year-old Han Chinese boy presented with abdominal pain and vomiting. The patient had tenderness in the right lower quadrant of the abdomen. Preoperative imaging suggested Meckel's diverticulum, but intraoperative exploration revealed both Meckel's diverticulum and intestinal duplication. Surgical resection and anastomosis were performed, and histopathology confirmed the diagnosis. The patient was discharged 7 days after surgery.

Conclusion Meckel's diverticulum combined with intestinal duplication malformations is rare, and intraoperative exploration is essential in the diagnosis and treatment of surgical disease.

Keywords Case report, Hidden diseases, Intestinal duplication, Intraoperative exploration, Meckel's diverticulum

Introduction

Meckel's diverticulum (MD) is a rare congenital anomaly that occurs when the omphalomesenteric duct fails to obliterate during fetal development. This results in a persistent connection between the ileum and the umbilical cord, leading to a diverticulum that can cause various complications. Intestinal duplication (ID), in contrast, is a rare congenital anomaly characterized by the presence of a duplicate segment of the bowel that is connected to the main intestine. This case report presents a rare instance of MD complicated with ID. Intraoperative exploration plays a crucial role in identifying unexpected anomalies that may not be detected by preoperative imaging. This case highlights the importance of thorough surgical exploration in diagnosing complex congenital malformations.

Case report

A 5-year-old Han Chinese boy was admitted to the emergency department with a 1-day history of persistent abdominal pain and vomiting. He had no fever, abdominal distension, or hematochezia. The patient had a prior history of intermittent right-sided abdominal pain and constipation, which had previously resolved

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spontaneously without the need for analgesics. However, this time, the right-sided abdominal pain became persistent and did not improve significantly with rest, leading him to seek medical attention. Upon physical examination, tenderness was noted in the right lower quadrant of the abdomen, but there was no rebound tenderness or muscle guarding.

Initial investigations revealed a white blood cell (WBC) count of 7.9×10^9/L, with a neutrophil predominance. B-ultrasound identified a 27 mm×19 mm×15 mm anechoic mass with clear boundaries and muscular echoes in the right lower quadrant of the abdomen (Fig. 1A), suggestive of Meckel's diverticulum. A subsequent computed tomography (CT) scan confirmed a 31 mm×26 mm×12 mm diverticulum located in the right lower quadrant arising from the ileum (Fig. 1B).

Laparoscopic exploration was performed to evaluate the abdominal findings. Intraoperatively, MD was identified 70 cm from the ileocecal valve. Additionally, an intestinal duplication ID was discovered 2 cm proximal to the ileocecal region, which had not been detected during preoperative imaging (Fig. 2A, B). Surgical resection of the affected bowel segment, including both the MD and ID, was performed, followed by anastomosis of the remaining healthy bowel (Fig. 2C–E). Postoperatively, the patient's status was managed with fasting, antimicrobial therapy, and intravenous fluid support. Oral intake was gradually reintroduced as tolerated. The patient remained clinically stable throughout the postoperative course and was discharged on the seventh postoperative day without complications. At the 1-month follow-up visit, physical examination revealed no abdominal abnormalities, and ultrasonography showed no signs of recurrence. The patient reported complete resolution of symptoms and was tolerating a regular diet. Ethical committee approval was not required for this case report, as it involved retrospective analysis of anonymized data, and written informed consent was obtained from the patient's parents. Histopathological examination of the resected specimen confirmed the diagnosis of MD and ID, with no evidence of ectopic gastric mucosa (Fig. 3). Immunohistochemical analysis of the resected specimens revealed no significant differences in the expression of several transcription factors between the specimens (Fig. 4).

Discussion

MD is a rare congenital anomaly that affects approximately 1–3% of the population, the condition is more common in males, with a ratio of 3:1 [1]. Johann F Meckel, a famous anatomist, was the first to describe this condition in 1809. Typically, MD often occurs in the opposite mesenteric wall of ileum within 100 cm of ileocecal valve. It is caused by the failure of the omphalomesenteric duct to obliterate during fetal development, leading to a persistent connection between the ileum and the umbilical cord [2]. In normal embryology, the vitelline duct closes by the tenth week of gestation. If it persists, presentations include incidental Meckel's diverticulum, fibrous cord connecting the bowel to the anterior abdominal wall, persistent omphalenteric fistula, torsion, and intussusception. These presentations can cause various complications, such as intestinal obstruction, hemorrhage, intussusception, and infection [3]. Although most MD can be asymptomatic for life, about 50% of MD contains ectopic tissues, such as gastric mucosa and pancreatic tissue, which are important factors leading to complications and clinical symptoms of MD [4]. Most MD is exogenous, but some cases are inverted, which may affect clinical judgment, increase

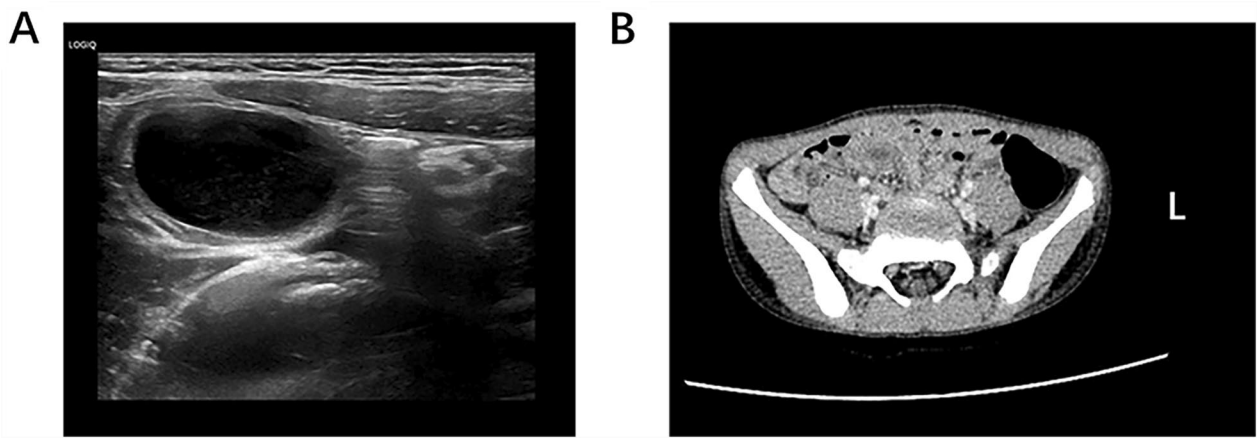

Fig. 1 The results of imaging examination of the patient. **A** B-ultrasound showed that there was no echo mass in the right lower abdomen. **B** Computed tomography showed a mass of low-density focus in the right lower abdomen

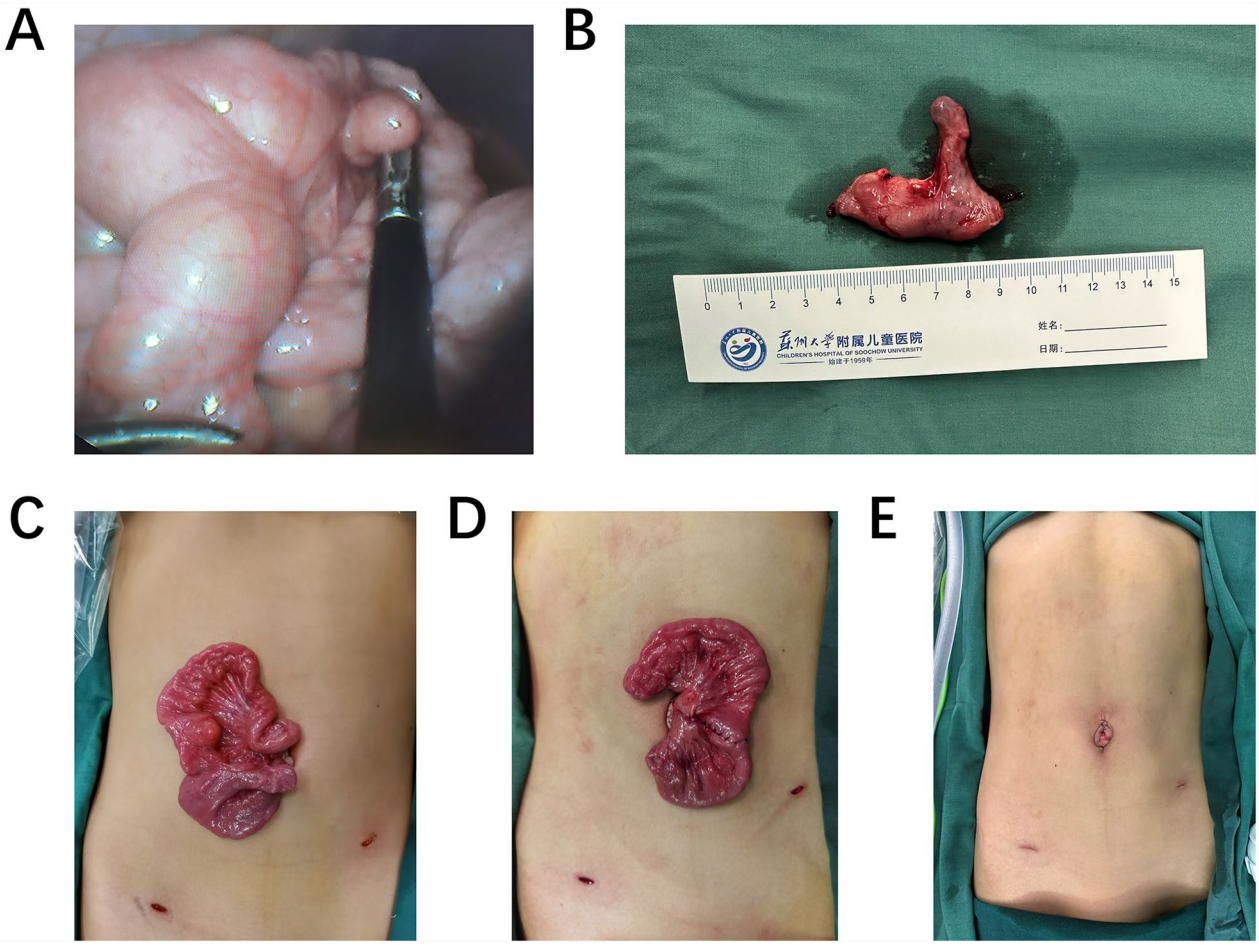

Fig. 2 Lesion intraoperately and postoperatively. **A** Laparoscopy revealed diseased bowel. **B** Postoperative dissociation of tissue. **C** Dragging out of the diseased bowel via the umbilicus. **D** Resection and repair of the diseased bowel. **E** Surgery completed

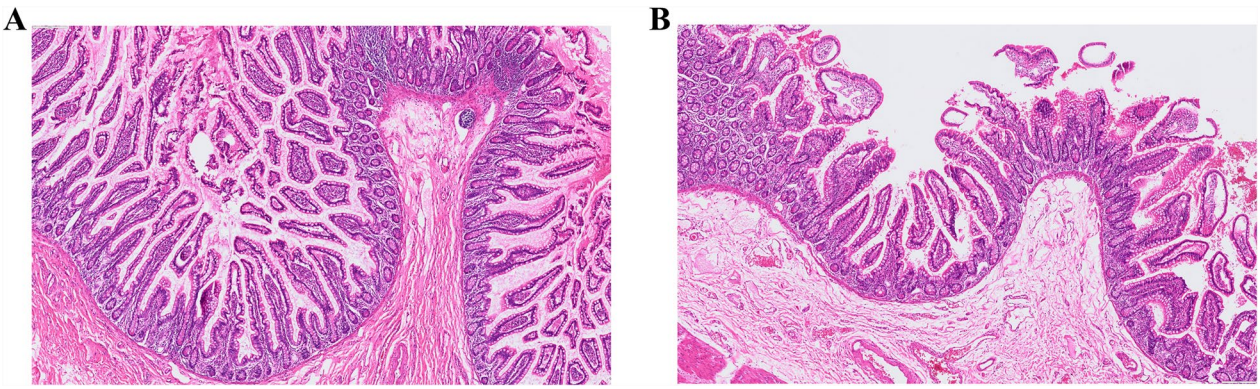

Fig. 3 Histological examination of the diseased bowel suggested no ectopic gastric mucosa or pancreatic tissue. **A** Pathological examination of Meckel's diverticulum. **B** Pathological examination of intestinal duplication malformations

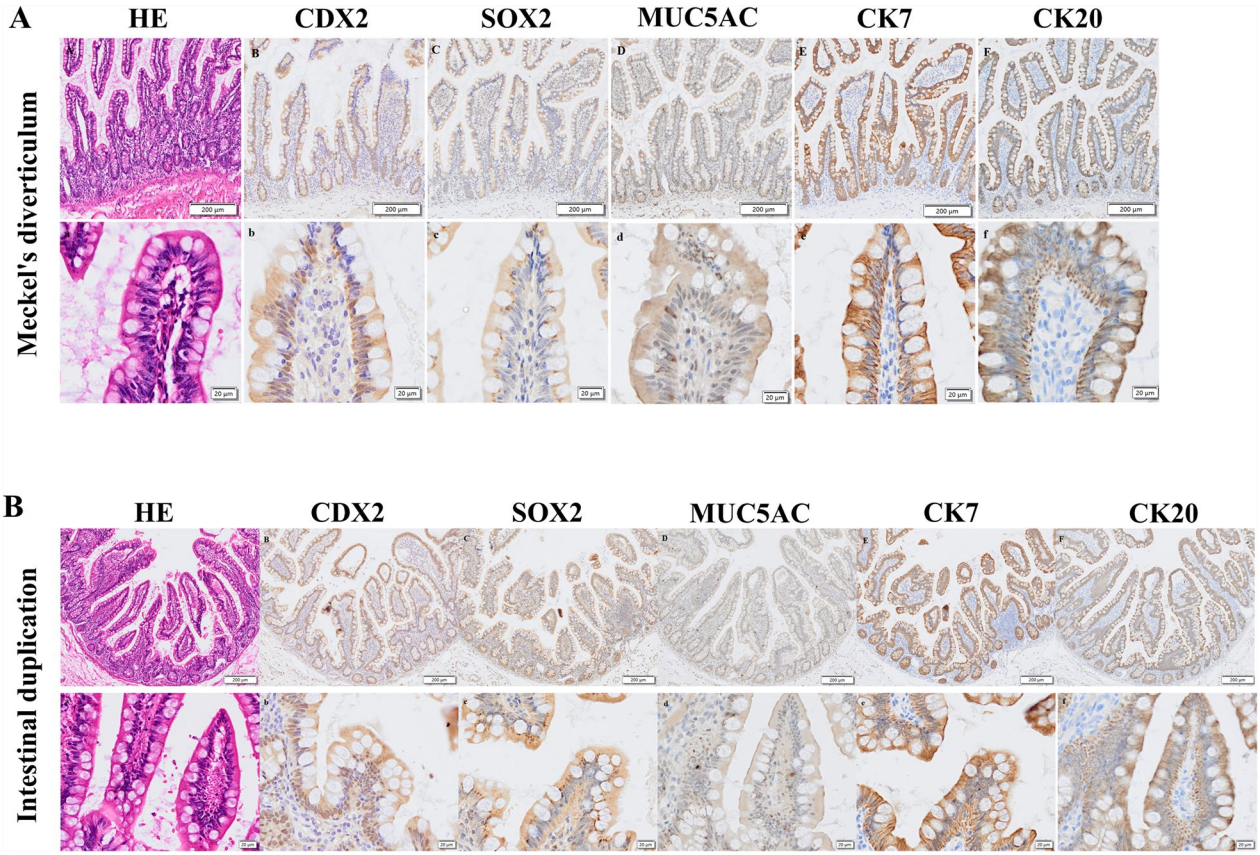

Fig. 4 Immunohistochemical analysis. **A** In the Meckel's diverticulum specimens, *CDX2*, *SOX2*, *MUC5AC*, *CK7*, and *CK20* were positive. **B** In the intestinal duplication specimens, *CDX2*, *SOX2*, *CK7*, and *CK20* were positive, while *MUC5AC* was weakly positive

the difficulty of operation, and delay the opportunity of operation [5].

Embryonic development involves many cellular processes, such as migration, proliferation, cycle, and apoptosis [6]. During the early development of the embryonic intestine, various transcription factors play critical roles. For instance, *SOX2* is primarily expressed in the anterior regions of the gut, such as the esophagus and stomach (proximal intestine), whereas *CDX2* is predominantly found in the posterior regions, including the small and large intestines [7]. Cytokeratins (CKs), which serve as intermediate filaments, are essential components of the intracellular cytoskeleton in epithelial cells [8]. Mucins, a group of high-molecular-weight glycoproteins, are secreted by epithelial cells; among these, *MUC5AC* is specifically associated with gastric mucosa [9]. In this case, hematoxylin and eosin (HE) staining did not reveal any obvious ectopic tissue within the Meckel's diverticulum. To further investigate the possibility of ectopic tissue presence, immunohistochemical staining was performed using specific markers associated with gastric mucosa (for example, *MUC5AC*) and intestinal differentiation (for example, *SOX2* and *CDX2*). However, the immunohistochemical analysis showed no positive staining for these markers, suggesting the absence of detectable ectopic gastric or pancreatic tissues within the diverticulum. This finding is consistent with the patient's lack of clinical symptoms related to ectopic tissue activity, such as gastrointestinal bleeding or ulceration. It also supports the hypothesis that the diverticulum in this case was primarily composed of normal intestinal mucosa without significant ectopic elements. In contrast to previous reports, our case is unique in the absence of ectopic tissue. This discrepancy may be attributed to variations in embryonic development or the specific genetic and molecular mechanisms underlying the formation of the diverticulum. While the absence of ectopic tissue reduces the risk of complications, close clinical follow-up remains important to monitor for any potential late-onset symptoms. In addition to genes, many factors, such as long non-coding RNA, are involved in regulating the expression of protein [10], which in turn affects the physiological or pathological process. Previous

studies have confirmed that *ZFAS1* and *MIR31HG* are involved in the pathogenesis of Hirschsprung's disease [11, 12]. No related investigations confirmed the role of non-coding RNA in the development of MD. Future research should focus on elucidating how these non-coding RNAs regulate key genes involved in embryonic development, potentially influencing cellular processes such as migration, proliferation, and apoptosis, which may contribute to the development of these congenital anomalies.

Alimentary tract duplication (ATD) is a rare congenital malformation, which refers to a spherical or tubular cavity structure located on the mesenteric side of digestive tract [13]. Ladd popularized the term "intestinal duplication" in 1937, and pointed out that IDs share blood supply with normal tissues [14]. ATD, which is usually found in the jejunum and ileum, can occur in any part of digestive tract, and the incidence rate is about 1/4500 [15]. According to the existing literature, the shape and size of digestive tract duplication vary greatly; this can be divided into four types in pathology: cystic type, tubular type, thoracic digestive tract duplication, and cervical digestive tract duplication [16, 17]. ATD in the oral cavity has also been reported, but is extremely rare [18]. Clinical manifestations include intestinal obstruction, abdominal pain, and infection. Since the ectopic tissues in the duplication, bleeding may also exist [19]. Duplication of digestive tract can be accompanied by duplication of other organs, such as double uterus and double vagina, with corresponding symptoms [20].

Surgery is an effective treatment option. The surgical method varies according to the site of the deformity. A site that is easy to remove is usually done by partial excision of the digestive tract and anastomosis; special sites that are difficult to excise should be drained and the mucosa stripped. Preoperative examination revealed the presence of MD, but intraoperative exploration again confirmed the presence of intestinal duplication of the malformation, avoiding the possibility of missing a microscopic lesion. This shows that intraoperative exploration is essential in the diagnosis and treatment of surgical diseases, as it can compensate for the omission of preoperative examinations and identify lesions that cannot be easily detected by preoperative examinations. MD complicated with ID is a rare and challenging surgical condition. Early diagnosis and surgical intervention are crucial in preventing complications and improving outcomes. Future research should integrate molecular biology, genetics, and clinical medicine to elucidate the underlying mechanisms of these conditions and develop novel diagnostic and therapeutic strategies. This case report highlights the importance of considering this rare anomaly in the differential diagnosis of abdominal pain and distension in children.

Conclusions

The co-occurrence of Meckel's diverticulum and intestinal duplication malformations is exceptionally rare. This case underscores the critical role of intraoperative exploration in diagnosing complex congenital anomalies, particularly when preoperative imaging fails to provide definitive findings. Clinicians should maintain a high index of suspicion for hidden anomalies, especially in pediatric patients presenting with persistent or unexplained abdominal symptoms.

Acknowledgements
The authors thank Prof. Zhicheng Gu from the Department of Pediatrics, Soochow University Children's Hospital, for his support of all the authors in their work.

Author contributions
PC, RZ, XX, and TZ performed diagnostic tests and collected data. PC and RZ wrote the first draft of the article. HZ, YJ, QW, MZ, and XZ took care of the patient. JC, JZ, SH, and ZZ raised critical comments on the article. PC and ZZ designed the case report. All authors have read and approved the manuscript.

Funding
This study was supported by the Natural Science Foundation of China (NSFC 82100534), Suzhou Science Foundation (SYW2024106), Key Laboratory of Structural Deformities in Children of Suzhou (NO. SZS2022018), and Gusu Health Talents Project (GSWS2022060).

Data availability
All data relevant to the study are included in the article or uploaded as supplemental information.

Declarations

Ethics approval and consent to participate
The study was approved by the Children's Hospital of Soochow University Ethics Committee. Written informed consent was obtained from the patient's parents for the publication of this case report and any accompanying images.

Consent for publication
Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors declare that they have no competing interests.

Received: 11 November 2024 Accepted: 1 April 2025
Published online: 18 May 2025

References

1. Yan P, Jiang S. Tc-99m scan for pediatric bleeding Meckel diverticulum: a systematic review and meta-analysis. J Pediatr (Rio J). 2023;99(5):425–31.
2. El Hajra MI, Calvo M, Martinez-Porras JL, Gomez-Pimpollo Garcia L, Rodriguez JL, Leon C, *et al*. Inverted Meckel's diverticulum diagnosed using capsule endoscopy: a case report. World J Gastroenterol. 2021;27(36):6154–60.

3. Rashid OM, Ku JK, Nagahashi M, Yamada A, Takabe K. Inverted Meckel's diverticulum as a cause of occult lower gastrointestinal hemorrhage. World J Gastroenterol. 2012;18(42):6155–9.
4. Keese D, Rolle U, Gfroerer S, Fiegel H. Symptomatic Meckel's diverticulum in pediatric patients—case reports and systematic review of the literature. Front Pediatr. 2019;7:267.
5. Jia X, Ma T, Liu G, Zuo X. Inverted Meckel diverticulum diagnosed, treated, and followed up by enteroscopy. Am J Gastroenterol. 2023;118(6):927.
6. Piprek RP, Kloc M, Mizia P, Kubiak JZ. The central role of cadherins in gonad development, reproduction, and fertility. Int J Mol Sci. 2020;21(21):8264.
7. Raghoebir L, Biermann K, Buscop-van Kempen M, Wijnen RM, Tibboel D, Smits R, *et al*. Disturbed balance between SOX2 and CDX2 in human vitelline duct anomalies and intestinal duplications. Virchows Arch. 2013;462(5):515–22.
8. Herrmann H, Bär H, Kreplak L, Strelkov SV, Aebi U. Intermediate filaments: from cell architecture to nanomechanics. Nat Rev Mol Cell Biol. 2007;8(7):562–73.
9. Kufe DW. Mucins in cancer: function, prognosis and therapy. Nat Rev Cancer. 2009;9(12):874–85.
10. Soutschek M, Schratt G. Non-coding RNA in the wiring and remodeling of neural circuits. Neuron. 2023;111(14):2140–54.
11. Wang Y, Cai P, Wang J. Clinical significance and biological effect of ZFAS1 in Hirschsprung's disease and preliminary exploration of its underlying mechanisms using integrated bioinformatics analysis. Ir J Med Sci. 2022;191(6):2669–75.
12. Cai P, Li H, Huo W, Zhu H, Xu C, Zang R, *et al*. Aberrant expression of LncRNA-MIR31HG regulates cell migration and proliferation by affecting miR-31 and miR-31* in Hirschsprung's disease. J Cell Biochem. 2018;119(10):8195–203.
13. Morris G, Kennedy A Jr. Small bowel congenital anomalies: a review and update. Surg Clin North Am. 2022;102(5):821–35.
14. Tanabe ID, DiTomaso A, Pinkas H, Pencev D. Massive GI hemorrhage from an ileal duplication cyst in an adult. Am J Gastroenterol. 1995;90(3):504–5.
15. Li GB, Han JG, Wang ZJ, Zhai ZW, Tao Y. Successful management of tubular colonic duplication using a laparoscopic approach: a case report and review of the literature. World J Clin Cases. 2020;8(15):3291–8.
16. Sanguesa Nebot C, Llorens Salvador R, Carazo Palacios E, Pico Aliaga S, Ibanez PV. Enteric duplication cysts in children: varied presentations, varied imaging findings. Insights Imag. 2018;9(6):1097–106.
17. Karnak I, Ocal T, Senocak ME, Tanyel FC, Buyukpamukcu N. Alimentary tract duplications in children: report of 26 years' experience. Turk J Pediatr. 2000;42(2):118–25.
18. Delay A, Morand B, Hacini L, Lewandowski M, Baudin F, Bachelet JT. "Oris duplicatio"-duplication of the oral cavity a rare malformation in a 10-month-old child: state of the science and management. J Craniofac Surg. 2022;33(8):2548–50.
19. Chen JJ, Lee HC, Yeung CY, Chan WT, Jiang CB, Sheu JC. Meta-analysis: the clinical features of the duodenal duplication cyst. J Pediatr Surg. 2010;45(8):1598–606.
20. Okur H, Keskin E, Zorludemir U, Olcay I. Tubular duplication of the hindgut with genitourinary anomalies. J Pediatr Surg. 1992;27(9):1239–40.

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