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Case Report

Meckel's diverticulum leading to ileo-ileal intussusception ☆☆☆

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ABSTRACT

Intussusception secondary to Meckel's diverticulum is a rare entity in adults and hence, can be a challenging to accurately diagnose preoperatively. This case illustrates the clinical, imaging, operative, and histologic manifestations of a Meckel's diverticulum leading to a long segment ileo-ileal intussusception in an adult female patient presenting with symptoms of small bowel obstruction.

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Introduction

Intussusception is the process of telescoping of a proximal portion of the bowel into the distal segment leading to complications such as ischemia and bowel obstruction [1,2]. This pathology is common in children but rare in adults, accounting for only 5% of the total incidence [1]. While most cases of pediatric intussusception are idiopathic, those of adult are usually associated with a malignant process [2,3].

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract, arising from the persistent remnant of the omphalomesenteric duct beyond fetal development [4]. MD tends to become symptomatic during childhood with bleeding, obstruction, or inflammation [5,6]. In adults, however it was reported that only 1 out of 300-400 of acute abdomens are caused by a complicated MD [7]. Because of the rarity of the disease in adults and its overlapping

symptoms with other inflammatory, obstructive and neoplastic processes, the diagnosis is often overlooked and can almost always be definitively proven intraoperatively.

Case presentation

A 38-year-old female patient with no known past medical or surgical history presented to the emergency room with diffuse abdominal pain for 1 week. The pain was intermittent, cramp-like, and associated with bloody stools and bilious emesis. On physical examination, the patient was afebrile, anicteric with a normal cardiovascular, and respiratory examination. The abdomen was distended with diffuse tenderness to palpation in all 4 quadrants. Initial laboratory workup demonstrated leukocytosis with a white blood count of 11,700 (reference range 4300-11000/ μ L) and elevated lipase of 798 (reference range

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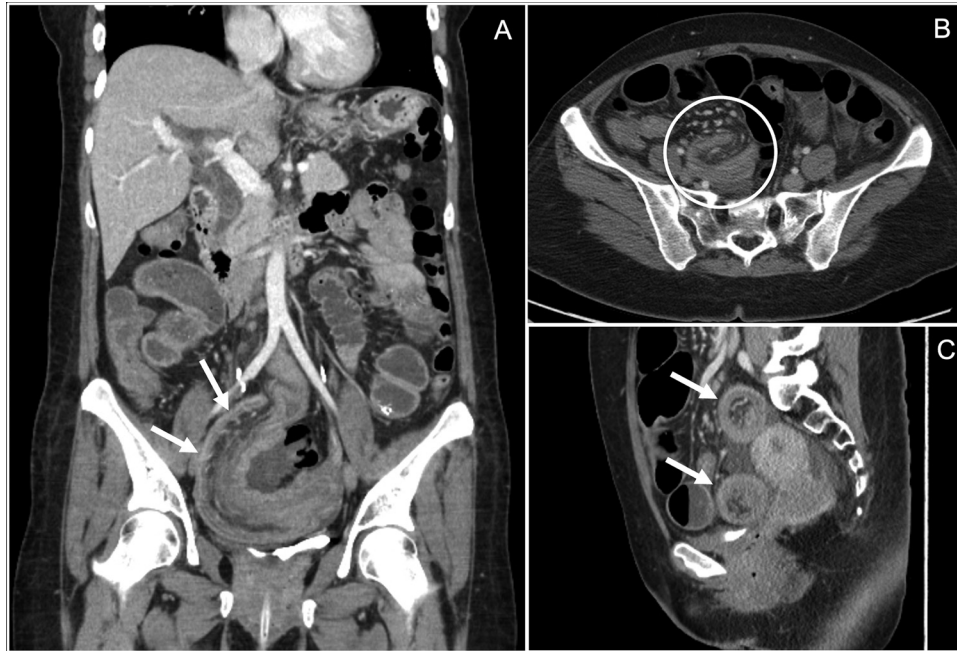


Fig. 1 – Coronal (A), axial (B), and sagittal (C) images from contrast enhanced CT of the abdomen and pelvis showing a long segment ileo-ileal intussusception in the distal small bowel. Telescoping of proximal ileum into a distal ileal segment is seen in a curvilinear or sausage shape (arrows in A). No discrete soft tissue mass or lead point is identified.

0–220 U/L), but normal electrolytes, and hepatic functions. Her pregnancy test was negative.

A contrast-enhanced computed tomography (CT) of the abdomen and pelvis was obtained for further evaluation. The imaging demonstrated dilatation of mid- to distal small bowel, proximal to a long segment intussusception in the distal ileum. Focal distention was noted along the distal aspect of the intussusception, but no clear soft tissue mass was identified. There was no evidence of peripancreatic inflammatory changes (Fig. 1).

An exploratory laparotomy was performed. An extensive ileo-ileal intussusception was seen about 80% of which was reduced by applying manual pressure on the intussusciptens. However, the entire length could not be reduced, and a segmental small bowel resection was performed, followed by an end-to-end anastomosis. The resected specimen was examined in the operating room, and a Meckel's diverticulum was identified. Surgical pathology confirmed the diagnosis of a Meckel's diverticulum with a small bowel intussusception. No malignancy or ectopic gastric mucosa was identified (Fig. 2). Postoperatively, the patient recovered well, and was subsequently discharged.

Discussion

Intussusceptions in adults account for only 5% of the total incidence [1]. The condition is typically classified by location and etiology. Intussusception can also be further divided by the presence of a lead point. Cases without lead point are often transient and spontaneously resolving. No bowel obstruction

is present and hence, treatments are usually unnecessary. Intussusception with lead point, however, is persistent and/or recurrent with the presence of bowel obstruction and as a result, usually requires surgical intervention [8]. In adults, the most common cause of intussusception is a structural lesion. A single-center review reports malignancy be the most common cause for a colonic intussusception while a benign process commonly causes a small bowel intussusception [9].

Meckel's diverticulum is a normal variant in 2% of the population. Embryologically, the diverticulum is the remnant of the vitelline duct, located on the antimesenteric border of the ileum within about 100 cm of the terminal ileum. Complications of MD often occur in childhood and adolescence and less in adults. While hemorrhage is the most common presenting symptom in children, symptomatic MD in adults tend to cause bowel obstruction (36.5%). After 40 years of age, however, tumor is the most common cause of MD complications [7]. Other medical problems arising from MD include intussusception which often first presents as obstruction, inflammation or diverticulitis, perforation, hemorrhage, neoplasm, and fistula [10]. Rare complications, such as regional enteritis, hernia, enterolith formation and calcification, tuberculosis, foreign bodies, and parasites have also been reported. It is estimated that the total lifetime risk of developing MD complication was 4% at 16 years of age, which declined to zero in old age [11]. In adult patients, MD has a complication rate of 0.03% per year [11]. Therefore, MD is usually overlooked on the differential of an acute abdomen in adults.

Intussusception caused by MD is uncommon and has been estimated to occur in 4% of all cases of intestinal obstruction attributed to intussusception [12]. Pre-operative diagnosis of intussusception secondary to MD has remained a challenge

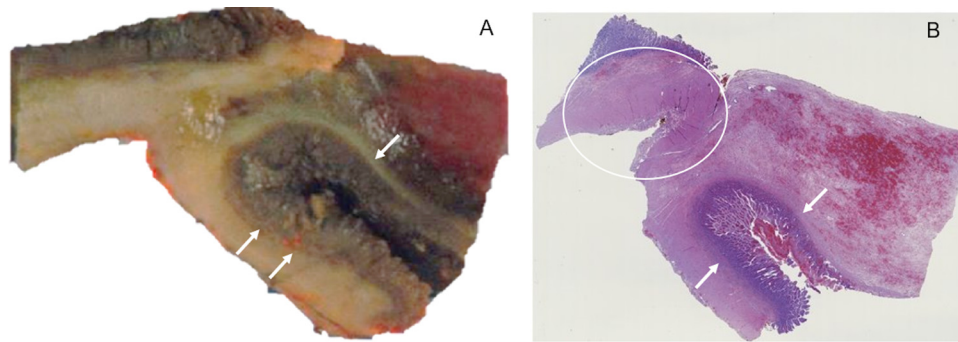


Fig. 2 – (A) Gross images of the surgical specimen show a Meckel's diverticulum (arrows), the intussusceptum, adherent to the surrounding small bowel wall (intussusciens). (B) Microscopic views (4x) showing Meckel's diverticulum (arrows) and adjacent small bowel mucosa (circle).

and hence, requires comprehensive data from clinical, pathologic, radiological and surgical findings. Clinically, patients with intussusception often present with symptoms that overlap with other types of bowel obstruction, including emesis, acute abdominal pain, tenderness, and distention. Other classical findings include fever, slightly elevated leukocyte count, bleeding, and history of recurrent bouts of abdominal pain [13].

Radiologically, plain film is sensitive in picking up intestinal obstruction secondary to MD which often shows dilated loops of small bowel with multiple air-fluid levels, bowel perforation or enteroliths [10]. However, this modality is low yield in picking up MD as the direct cause of an acute abdomen. Fluoroscopically, MD appears as a saccular outpouching from the antimesenteric border of the distal ileum or a filling defect when inverted [14]. Ultrasound is another modality that can aid in the diagnosis of MD intussusception. The inverted diverticulum often shows as a “double target” sign, also known as the doughnut or bull's eye sign. CT, however, is the more definitive diagnostic tool for bowel obstruction with a better visualized target lesion. The finding represents the stranded bowel viewed in cross section with bowel wall thickening and mesenteric stranding. It is also sensitive in diagnosing leadpoint vs non-leadpoint intussusception, which helps guide the appropriate management. Other lesions, such as enteroliths, and other less heavily calcified stones can also be localized on CT scan [14]. Nevertheless, the scan often does not specifically pinpoint MD as the leading point of intussusception. To further narrow the differential diagnosis, Meckel's scan has been heavily utilized due to the uptake of Technetium-99m pertechnetate by heterotopic gastric mucosa. The sensitivity of the study is enhanced by pentagastrin, H2 receptor antagonist, and glucagon. However, there are also several causes false positives due to duodenal, and jejunal duplication cysts containing gastric mucosa [14]. False negatives can also occur due to lack of gastric mucosa or residual contrast medium within the abdomen from a previous radiologic study.

Pathologically, MD is a true diverticulum that contains all layers of intestinal wall. Heterotopic tissues include gastric, pancreatic, duodenal, jejunal, colonic, rectal, and endometrial

mucosa are frequently found in Meckel's diverticulum [15]. Neoplasm can also develop in the ectopic tissues with neuroendocrine being the most common tumor [16]. Bowel reduction, diverticulectomy, and/or bowel resection are required to relieve intussusception caused by MD. Bowel resection is indicated in case on transmural ischemia [17]. With the increasing incidental findings of MD on imaging and intraoperatively, there have been ongoing controversies regarding the management strategies of these cases. In asymptomatic patients with incidental MD on imaging, resection is generally not indicated [18]. Elective surgery, however, is recommended in a selective approach when MD is found intraoperatively. Surgical complications, including deaths have been reported [7]. Incidental removal of MD has been reported to have a morbidity and mortality rates of 3.5% and 0.2%, respectively. In symptomatic cases, morbidity is increased to 5.25%, and patients bear a 1.5% risk of mortality [7]. As a result, great consideration should be taken when deciding the appropriate management in otherwise young and healthy patients. Factors, such as male sex, age <40, anesthetic risks, the type of surgery, the size, location, length (>2cm) and morphology (narrow base of implantation) of the diverticulum, and presence of a palpable mass can indicate higher risk of complications later in life, and hence, favor surgical removal of an asymptomatic MD [7].

Conclusion

Intussusception in adults is usually associated with an underlying structural pathology, especially non-transient or recurrent cases. Therefore, it is important to thoroughly evaluate the region of the abnormality on imaging to look for a lesion that may serve as the lead point. Because it can be challenging to pinpoint MD as the pathologic lesion, surgical resection and pathology can provide a definitive diagnosis. While both a Meckel's diverticulum and intussusception are rare in adults, it is important for radiologists to be cognizant of their clinical and radiological picture, especially when other more common differential diagnoses have been ruled out.

Authorship

The authors declare that this is their original work and they all approve the content of this manuscript. They confirm that this manuscript has not been published previously, in any language, in whole or in part, and is not currently under consideration elsewhere.

Ethical clearance

This project did not involve any research and no ethical clearance was required.

Patient consent

A written informed consent was obtained from the patient for the publication of this case report.

REFERENCES

- [1] Marinis A, Yiallourou A, Samanides L, Dafnios N, Anastasopoulos G, Vassiliou I, et al. Intussusception of the bowel in adults: a review. *World J Gastroenterol* 2009;15(4):407–11.
- [2] Honjo H, Mike M, Kusanagi H, Kano N. Adult intussusception: a retrospective review. *World J Surg* 2015;39(1):134–8.
- [3] Lu T, Chng YM. Adult intussusception. *Perm J* 2015;19(1):79–81.
- [4] Sagar J, Kumar V, Shah DK. Meckel's diverticulum: a systematic review. *J R Soc Med* 2006;99(10):501–5.
- [5] Hansen CC, Søreide K. Systematic review of epidemiology, presentation, and management of Meckel's diverticulum in the 21st century. *Medicine (Baltimore)* 2018;97(35):e12154.
- [6] Chen JJ, Lee HC, Yeung CY, Chan WT, Jiang CB, Sheu JC, et al. Meckel's diverticulum: factors associated with clinical manifestations. *ISRN Gastroenterol* 2014;2014:390869.
- [7] Groebli Y, Bertin D, Morel P. Meckel's diverticulum in adults: retrospective analysis of 119 cases and historical review. *Eur J Surg* 2001;167(7):518–24.
- [8] Kim YH, Blake MA, Harisinghani MG, Archer-Arroyo K, Hahn PF, Pitman MB, et al. Adult intestinal intussusception: CT appearances and identification of a causative lead point. *Radiographics* 2006;26(3):733–44.
- [9] Huang WS, Changchien CS, Lu SN. Adult intussusception: a 12-year experience, with emphasis on etiology and analysis of risk factors. *Chang Gung Med J* 2000;23(5):284–90.
- [10] Dumper J, Mackenzie S, Mitchell P, Sutherland F, Quan ML, Mew D. Complications of Meckel's diverticula in adults. *Can J Surg* 2006;49(5):353–7.
- [11] Leijonmarck CE, Bonman-Sandelin K, Frisell J, Räf L. Meckel's diverticulum in the adult. *Br J Surg* 1986;73(2):146–9.
- [12] Agha FP. Intussusception in adults. *AJR Am J Roentgenol* 1986;146(3):527–31.
- [13] Intussusception of the small intestine with special reference to meckel's diverticulum as a causative factor. *N Engl J Med* 1928;199(10):453–6.
- [14] Thurley PD, Halliday KE, Somers JM, Al-Daraji WI, Ilyas M, Broderick NJ. Radiological features of Meckel's diverticulum and its complications. *Clin Radiol* 2009;64(2):109–18.
- [15] DeBartolo HM, van Heerden JA. Meckel's diverticulum. *Ann Surg* 1976;183(1):30–3.
- [16] Kabir SA, Raza SA, Kabir SI. Malignant neoplasms of Meckel's diverticulum; an evidence based review. *Ann Med Surg (Lond)* 2019;43:75–81.
- [17] Sioka E, Christodoulidis G, Garoufalis G, Zacharoulis D. Inverted Meckel's diverticulum manifested as adult intussusception: age does not matter. *World J Gastrointest Surg* 2011;3(8):123–7.
- [18] Zani A, Eaton S, Rees CM, Pierro A. Incidentally detected Meckel diverticulum: to resect or not to resect? *Ann Surg* 2008;247(2):276–81.