

Intussusception secondary to a Meckel's Diverticulum in an adolescent: a case report

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Abstract

Background: Intussusception is a common cause of abdominal pain and a leading cause of bowel obstruction in young children

Case presentation: Herein, we report a 16-year-old male who appeared in the emergency department with a history of abdominal pain localized in the right lower abdomen. Clinical examination revealed a positive McBurney sign. Laboratory studies revealed elevated white blood cells, neutrophils, and C-reactive protein, while abdominal ultrasound disclosed no pathology in the abdomen or free fluid. The patient underwent an open appendectomy due to the high suspicion of acute appendicitis. An ileo-ileac intussusception was found at the operation with a Meckel's diverticulum as the lead point, which was resected.

Conclusion: Intussusception is common in children and rare in adults. Its diagnosis is challenging due to non-specific symptoms. Therefore, general surgeons must be familiar with the epidemiology, etiology, diagnosis, and treatment of pediatric and adult intussusception.

Keywords: Intussusception; Meckel's diverticulum; Pediatric; Target sign; Pseudo-kidney sign; Enema reduction; Resection.

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I. Introduction

Intussusception is a leading cause of abdominal pain and bowel obstruction in children. It is common in children and rare in adults (1). The estimated frequency is 26-38 cases per 100,000 live births in the first three years of life (2). In the pediatric population, the majority of cases are idiopathic without an identifiable lead point. It is estimated that 95% of cases in infants and toddlers are idiopathic, whereas, in about 5% of cases, a lead point is identified. The likelihood of identifiable causes of intussusception rises with increasing age (3).

Meckel's diverticulum is the most typical lead point. It is a true congenital intestinal diverticulum located 7 to 200 cm away from the ileocecal valve, typically on the anti-mesenteric border and rarely on the mesenteric side. It is 0.4 to 11 cm long with a diameter of 0.3 to 7 cm (4).

Abdominal ultrasound is the imaging modality of choice for the diagnosis of pediatric intussusception. Nevertheless, abdominal ultrasound detects pathological lead points in less than 30% of cases (5). Reduction by ultrasound-guided or fluoroscopic pneumatic or hydrostatic enema is the preferred treatment. Surgery is indicated when enema reduction is unsuccessful, signs of bowel necrosis or perforation are present, or a pathological lead point is the cause of intussusception. In the case of an intussuscepted Meckel's diverticulum, resection and subsequent anastomosis are recommended. Here we describe the case of a 16-year-old male who suffered ileo-ileac intussusception, and he was misdiagnosed as acute appendicitis (5, 6).

II. Case Presentation

A 16-year-old male presented to the emergency department with a history of abdominal pain localized in the right lower abdomen the previous 8 hours. The pain was described as cramping and constant with no radiation, associated with nausea and vomiting. There was no history of fever, diarrhea, constipation, or urinary

complaints. The patient visited the emergency department of our hospital four days ago with a sudden onset abdominal pain located in the right lower abdomen with no other symptoms. Laboratory findings were within normal ranges, and abdominal ultrasound revealed no pathology in the abdomen or free fluid (Figure 1). The pain was relieved with acetaminophen, and the patient was discharged after a few hours. The patient reported no significant previous medical or surgical history.

Abdominal examination disclosed a soft, non-distended abdomen, with deep tenderness at McBurney point and no evidence of peritonism. Laboratory studies revealed elevated white blood cells (12.8 K/UI), neutrophils (84%), and C-reactive protein (3.9 mg/dl). Abdominal ultrasound showed no free fluid, while the abdominal organs were normal (Figure 2). The appendix, however, was not visualized. The only finding was probe tenderness in the right iliac fossa. Unfortunately, the computed tomography was out of order.

The surgical team was consulted, and the patient underwent an open appendectomy due to a high clinical suspicion of acute appendicitis. Nevertheless, the appendix was identified, exposed, and noticed to be normal. An ileo-ileal intussusception was found with a Meckel's diverticulum as the lead point, which was resected. The patient recovered uneventfully, and he was discharged after four days.

III. DISCUSSION

Intussusception is a common cause of abdominal pain and a leading cause of bowel obstruction in young children. It is defined as the invagination of a proximal bowel segment (intussusceptum) into a more distal one (intussusciptens) (1). Its incidence varies from 26 to 38 cases per 100,000 live births in the first three years of life. Children younger than two years are more vulnerable to intussusception. It is reported that 60% of cases occur in children younger than one year, and 80-90% in children younger than two years, with a male predominance. However, intussusception should be considered in children at any age group as 10% of cases happen in children older than five years, and 3-4% in children older than ten years (2, 6, 7).

It is categorized as ileoileal, ileoileocolic, ileocolic (the most common type, more than 80%), and colocolic based on the involved bowel segments (1, 6). In the pediatric population, the vast majority of cases are idiopathic without an identifiable lead point. It is estimated that 95% of cases in infants and toddlers are idiopathic, whereas, in about 5% of cases, a lead point is identified. The likelihood of identifiable causes of intussusception rises with increasing age. Specific lead points are more frequently encountered in children older than three years and almost always in adults. Meckel diverticulum is the most typical lead point. Other causes encompass polyps, duplication cyst, hamartomas, lymphoma, carcinoma due to juvenile polyposis syndromes, lymphangiectasias, and Henocho-Schonlein purpura (3, 8).

Meckel's diverticulum is a true congenital intestinal diverticulum comprised of all three coats of the intestinal wall. It is caused by the incomplete obliteration of the vitelline duct during the fifth week of fetal development. Meckel's diverticulum might contain ectopic tissue in 4.6-71% of cases. The gastric mucosa is the commonest ectopic tissue (50-60%), followed by pancreatic, duodenal, and colonic tissue (5-6%). The estimated frequency of Meckel's diverticulum in the general population is 0.3-2.9% with a male predominance (2:1). It is located 7 to 200 cm away from the ileocecal valve, typically on the anti-mesenteric border and rarely on the mesenteric side. It is 0.4 to 11 cm long with a diameter of 0.3 to 7 cm. Patients are usually asymptomatic, but they may develop abdominal pain, gastrointestinal bleeding, bowel obstruction, and diverticulitis with or without intestinal perforation. Many surgical textbooks describe the rule of two concerning Meckel's diverticulum: present in 2% of the population, located 2 feet from the ileocecal valve, 2 inches long, 2% of patients will develop complications over the course of their lives, typically before the age of 2 years (4, 9-11).

Bowel obstruction and consequent congestion and edema are developed when a bowel segment telescopes into an adjacent one. Eventually, the lymphatic and vascular flow is jeopardized, resulting in ischemia to the affected part, leading to bowel necrosis, perforation, and peritonitis (1, 6).

Clinical diagnosis of intussusception is very challenging as the manifestation is variable and non-specific. Besides, the symptoms of intussusception overlap with other acute abdominal diseases. Typically, patients develop abdominal pain and signs of bowel obstruction. Vomiting and stools mixed with blood and mucus can also be observed. Physical examination may reveal a palpable "sausage-shaped" mass. However, in less than half of the cases, the classical triad of intussusception consisting of colicky abdominal pain, bloody stool, and palpable abdominal mass (12, 13).

Imaging modalities are the critical element for the diagnosis of intussusception. Although plain abdominal x-rays are useful in detecting bowel obstruction and perforation, they are insensitive to the diagnosis of intussusception. At present, abdominal ultrasound is the gold standard for diagnosing intussusception in children as it has a high sensitivity of 98.5-100%, high specificity of 88-100%, and a high negative predictive value of 100% (5, 14). The classic feature is the target or doughnut sign in the transverse view and the pseudo-kidney sign in the longitudinal view. The target sign is brought about by the edematous intussusciptens forming an external ring around the centrally based intussusceptum, while the layers of intussusception form a pseudo-kidney sign. On Doppler images, the absence of blood flow within the intussusception is associated with bowel

ischemia and necrosis. Although the differentiation of idiopathic intussusception and intussusception due to a pathological lead point is feasible using ultrasound, pathological lead points are diagnosed by ultrasound in less than 30% of cases. Abdominal computed tomography (CT) and magnetic resonance imaging (MRI) can also be used to diagnose intussusception in children and identify pathological lead points. The exposure to ionizing radiation on CT and the availability of MRI in most hospitals limit their use to diagnose intussusception in children (1, 5, 6, 15).

In the pediatric population, intussusception requires reduction by ultrasound-guided or fluoroscopic pneumatic or hydrostatic enema, which is successful in 85-90% of cases. Surgery is indicated when unsuccessful enema reduction, bowel necrosis or perforation are present, or pathological lead points cause intussusception. In the case of intussuscepted Meckel's diverticulum, the treatment of choice is resection with subsequent anastomosis. The operation may be performed via a laparotomy (usually) or laparoscopically, depending on the skill and experience of the surgeon (5, 6).

IV. Conclusion

Pediatric intussusception is a common and usually benign condition. Its diagnosis is challenging due to the lack of specific symptoms. Moreover, it is impossible to differentiate idiopathic versus secondary intussusception due to a pathological lead point based on signs and symptoms. Abdominal ultrasound is the imaging modality of choice for the diagnosis of pediatric intussusception. Nevertheless, abdominal ultrasound detects pathological lead points in less than 30% of cases. Treatment of pediatric intussusception is conservative. Reduction by ultrasound-guided or fluoroscopic pneumatic or hydrostatic enema is successful in 85-90% of cases. Indications of surgery include unsuccessful enema reduction, signs of bowel necrosis or perforation, and the presence of pathological lead points as the cause of intussusception. If the pathological lead point is an intussuscepted Meckel's diverticulum, resection and subsequent anastomosis are recommended.

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References

- [1]. Edwards EA, Pigg N, Courtier J, Zapala MA, MacKenzie JD, Phelps AS. Intussusception: past, present and future. *PediatrRadiol*. 2017;47(9):1101-1108.
- [2]. Buettcher M, Baer G, Bonhoeffer J, Schaad UB, Heininger U. Three-year surveillance of intussusception in children in Switzerland. *Pediatrics*. 2007;120(3):473-480.
- [3]. Mandeville K, Chien M, Willyerd FA, Mandell G, Hostetler MA, Bulloch B. Intussusception: clinical presentations and imaging characteristics. *PediatrEmergCare*. 2012;28(9):842-844.
- [4]. Marsicovetere P, Ivatury SJ, White B, Holubar SD. Intestinal Intussusception: Etiology, Diagnosis, and Treatment. *ClinColonRectalSurg*. 2017;30(1):30-39.
- [5]. Gluckman S, Karpelowsky J, Webster AC, McGee RG. Management for intussusception in children. *CochraneDatabaseSystRev*. 2017;6(6):CD006476.
- [6]. Deigaard SB, Trap R. Intestinal duplikaturenvigtigdifferentiaaldiagnoseved invagination [Intestinal duplication--an important differential diagnosis to intussusception]. *UgeskrLaeger*. 2008;170(35):2708.
- [7]. 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.
- [8]. Keese D, Rolle U, Gfroerer S, Fiegel H. Symptomatic Meckel's Diverticulum in Pediatric Patients-Case Reports and Systematic Review of the Literature. *FrontPediatr*. 2019;7:267.
- [9]. Malik AA, Shams-ul-Bari, Wani KA, Khaja AR. Meckel's diverticulum-Revisited. *Saudi J Gastroenterol*. 2010;16(1):3-7.
- [10]. Sagar J, Kumar V, Shah DK. Meckel's diverticulum: a systematic review. *J R Soc Med*. 2006;99(10):501-505.
- [11]. Daneman A, Alton DJ. Intussusception. Issues and controversies related to diagnosis and reduction. *RadiolClin North Am*. 1996;34(4):743-756.

- [12]. Samad L, Marven S, El Bashir H et al. Prospective surveillance study of the management of intussusception in UK and Irish infants. *Br J Surg.* 2012;99(3):411-415.
- [13]. Fiegel H, Gfroerer S, Rolle U. Systematic review shows that pathological lead points are important and frequent in intussusception and are not limited to infants. *Acta Paediatr.* 2016;105(11):1275-1279.
- [14]. Carroll AG, Kavanagh RG, Ni Leidhin C, Cullinan NM, Lavelle LP, Malone DE. Comparative Effectiveness of Imaging Modalities for the Diagnosis and Treatment of Intussusception: A Critically Appraised Topic. *AcadRadiol.* 2017;24(5):521-529.
- [15]. Li XZ, Wang H, Song J, Liu Y, Lin YQ, Sun ZX. Ultrasonographic Diagnosis of Intussusception in Children: A Systematic Review and Meta-Analysis. *J UltrasoundMed.* 2021;40(6):1077-1084.

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