Meckel diverticulum in children

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Abstract

Meckel diverticulum represents one of the most common malformations of the digestive tract due to incomplete obliteration of the omphalomesenteric duct. It occurs in 2% of the population, and approximately 4% of all Meckel diverticula cause complications, such as obstruction, haemorrhage, or inflammation. These complications typically occur before the age of five years and frequently require rapid surgical intervention. Diagnosis of a complicated Meckel diverticulum is difficult and often only made during surgery. In this article, the most important diagnostic characteristics and surgical approaches are summarized.

Introduction

Meckel diverticulum (MD) is a vestigial remnant of the omphalomesenteric duct that connects the yolk sac to the gut of the developing embryo in order to provide nutrition until the placenta is formed. Normally, the duct will obliterate around 7 weeks of gestation. However, failure of closure can result in a wide spectrum of anomalies including intestinal-umbilical fistula, omphalomesenteric duct sinus or cyst, mesodiverticular fibrous bands, umbilical polyp, and most commonly the MD (Figure 1). It is typically located approximately 52 cm (range 7-200 cm) proximal from the ileocecal valve, on the anti-mesenteric border, is 3 cm (range 0,4-11 cm) in length and 1,6 cm (range 0,3-7 cm) in diameter (1).

The prevalence of MD is between 0,3% and 2,9% in the general population (based on both findings during surgery and autopsy) with an estimated lifetime incidence of complicated MD of 4,2% (1,2). MD often becomes symptomatic in the first decade of life. The prevalence of symptomatic disease decreases with age, more than half of all symptomatic patients are younger than 10 years old, some authors even say younger than 5 years old (1,3). In symptomatic patients, there is a male predominance of about 2:1 (range 1,3-7,5:1), especially in patients with a nonobstructive presentation (3-15). Excessive acid secretion in males by ectopic gastric mucosa in the MD could be the reason for predisposition to bleeding or inflammation, as is also seen for peptic ulcer disease in males (3). The occurrence of gastric mucosa is common in symptomatic MD (45-80% of all cases). Other ectopic tissue that is less often seen in MD are pancreatic, duodenal or colonic. All other symptomatic MDs have ileal lining of the mucosa without ectopic tissue.

Even though the majority of MD remains asymptomatic, they have the potential to present with severe complications. The possible complications can be categorized into three groups: bowel obstruction, intestinal haemorrhage, and inflammation with or without perforation. The type of complications will depend on the different characteristics of the MD such as mechanical features (length and position of the MD) and the presence of ectopic gastric tissue located in the diverticulum. Diagnosis of a complicated MD may be challenging, due to its nonspecific symptoms that can mimic various other acute abdominal emergencies. If left untreated, necrosis, perforation, peritonitis or severe anaemia can occur, resulting in life threatening situations. Mortality is rare but has been described in the literature in 2 paediatric cases (4,7). Thus, a quick and accurate diagnosis is essential for a good outcome. The aim of this article is to review the current knowledge on clinical presentation, diagnostic workup, histopathological features, and management in children.

Clinical presentation

The clinical manifestation of the MD is highly variable. The most common symptom in children is abdominal pain. This is present in half of the paediatric patients (range 32,4-71,4%) (3,4,12-15). The pain is often located in the right fossa or around the umbilicus, mimicking an appendicitis. Other abdominal symptoms are presented in table 1. Since the clinical picture is highly dependent on the underlying mechanism, we will describe the three most important complications more into depth.

Figure 1: *Examples of congenital anomalies of omphalomesenteric duct.*
1. Fibrous connection of the ileum to the umbilicus, 2. Intestinal-umbilical fistula, 3. Omphalomesenteric duct sinus, 4. Omphalomesenteric duct cyst, 5. Meckel diverticulum with Meckel's band, 6. Meckel diverticulum.
Adapted from Netterimages: Meckel's Diverticulum: Variants of Vitelline Duct Remnants.

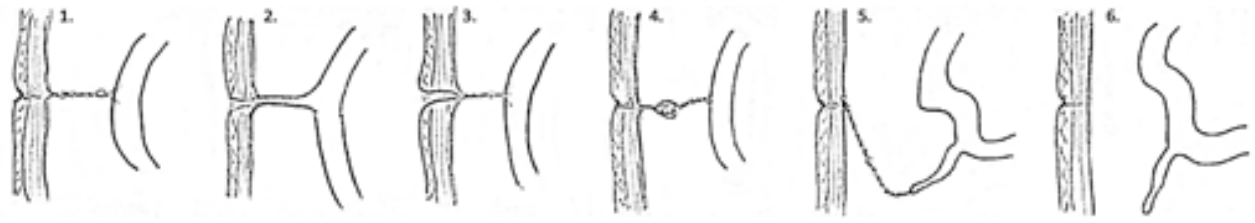


Table 1. Main presenting symptoms of Meckel's diverticulum in children

	Huang[3] (n = 100)	Keese[12] (n = 7)	Chen[4] (n = 233)	Francis[13] (n = 208)	Rho[14] (n = 34)	Tseng[15] (n = 45)
Rectal bleeding	50 (50%)	2 (29%)	116 (50%)	42 (20%)	12 (35%)	25 (56%)
Abdominal pain	57 (57%)	5 (71%)	129 (55%)	119 (57%)	11 (32%)	24 (53%)
Vomiting/nausea	53 (53%)	5 (71%)	104 (45%)	106 (51%)	3 (9%)	23 (51%)
Fever	25 (25%)	NR	59 (25%)	40 (19%)	NR	12 (27%)

NR: not reported

Intestinal obstruction

The most common complication of MD is intestinal obstruction, which is described in 41,5% of all symptomatic cases (range 14,3%-86,2%) (3-12,14,15).

MD can lead to an intestinal obstruction in several ways, intussusception being the principal presentation in the paediatric population (41,9%) (3,4,6-8,10-12,14). Ileo-ileal or ileo-colic intussusception can occur, where the MD will serve as a lead point. In addition, it is suggested that the higher nerve fiber density in the wall of the MD can increase the local peristalsis and induce the onset of intussusception (16). Abdominal pain and nausea or vomiting are the main presenting symptoms in patients with an intussusception. Rectal bleeding or the classic "red currant jelly" stool can be seen as late signs, suggesting bowel ischemia. Although intussusception is commonly idiopathic in children, in 5% of cases a pathologic lead point can be found, characterized by the inability to reduce the intussusception on contrast or air enema. In this group, a MD is the lead point in 37-80% of all cases (17-19).

The second most common cause of obstruction (20%) is volvulus around a Meckel band, which is a mesodiverticular band believed to be a remnant of a vitelline artery (7,10,14). Other types of obstruction can be due to kinking, knotting, strangulation and internal intestinal hernias through openings formed by a Meckel band or abnormal mesentery attached to the MD (Figure 2). A specific type of hernia which is rarely seen, is the Littre hernia, an internal hernia containing only the MD (4).

Intestinal haemorrhage

Intestinal bleeding from MD is commonly caused by ulceration of the small bowel due to the presence of ectopic gastric mucosa and its acid secretion. Gastric mucosa is present on microscopic histopathology in 45-80% of surgical specimens (4). Intestinal haemorrhage is seen in 37,2% (range 4,6-55,6%) of all complicated MDs (3-12,14,15). The bleeding typically has a dark colour and can be massive in volume, although more chronic forms have also been described with occult bleeding and anaemia (12). In about 50-75% of patients blood transfusion is required (3,4,11,14). Intestinal bleeding in MD patients can also be a presentation of intussusception, or a combination of ectopic gastric tissue and intussusception, in which cases the bleeding will be accompanied by abdominal pain.

Inflammation

The diverticulum can become inflamed due to bacterial overgrowth in the obstructed lumen, resulting in peritonitis due to diverticulitis with or without perforation. Peritonitis represents 20,8% (range 13-27,6%) of all symptomatic MD cases and is manifested by abdominal pain, nausea/vomiting and fever, mimicking the diagnosis of an appendicitis (3,5-11,14,15). Diverticulitis can lead to perforation which can be life threatening. This evolution is mentioned in four paediatric cohort series and it is probably seen in up to 15% of all symptomatic MDs (6,7,9,11). Finally, some case reports describe that the inflamed MD can present as a chronic form of ileitis that is misdiagnosed as isolated small bowel Crohn's disease (20).

Diagnostic approach

When there is clinical suspicion, radiological and nuclear imaging can be helpful in diagnosing complications from MD, and seldomly demonstrate MD as the cause. According to Daneman et al., the inflamed, haemorrhagic and the inverted, intussuscepted MD have a spectrum of features recognizable on ultrasound, CT and air enema in a significant number of children (44%) (21). Preoperative diagnostic rate in the study by Chen et al., with 233 symptomatic MD patients, was 24,89% (4). In the review by Rho et al., 57,6% of symptomatic MDs (20/34) were diagnosed by imaging techniques (14). The diagnosis of MD is in many cases made during exploratory surgery. Table 2 shows an overview of the most typical presentation forms and investigations likely to be helpful during diagnosis. In table 3, recommendations for practical use are suggested.

Diagnosing intestinal obstruction due to MD

As described earlier, there are different types of obstruction due to MD, intussusception being the most prevalent. Intussusception can be diagnosed on ultrasound in most cases, or, if necessary, on computed tomography (CT) scan (3). These investigations will not often reveal the MD as the lead point, as demonstrated by Huang et al., where the MD as the cause of intussusception was only confirmed during surgery in all of their 17 cases of intussusception. Furthermore, attempts for barium reduction failed or there was subsequent recurrence of intussusception (3). This should always raise concern for an obstruction related to a MD (22).

Other types of obstruction include volvulus, kinking, knotting, strangulation due to fibrous bands or herniation. Findings such as distended small bowel loops on plain radiograph or CT scan are helpful but not specific for MD. Lin et al. studied 102 paediatric patients with symptomatic MD, 48 patients underwent a CT scan, but only 3 patients revealed a suspected MD (8). In a smaller study by Olsen et al., MD was only mentioned in the CT report in five out of the 16 patients (31%) (23).

Diagnosing the bleeding MD

A pertechnate scintigraphy (Meckel scan) can identify ectopic gastric mucosa from a (bleeding) MD. Intravenous Technetium-99m pertechnate is taken up by the mucoid cells of gastric mucosa and subsequently excreted into the bowel lumen. Normal uptake is seen in the stomach, and to a lesser extent in the proximal small bowel. Subsequently, activity is seen in the bladder due to accumulation of radioactive urine after renal excretion (Figure 3).

A Meckel scan is the best noninvasive method for preoperative diagnosis of bleeding MD with high sensitivity (86-100%) and a variable specificity (22-100%). False negative results could be attributed to the "washout" phenomenon. Excessive bleeding or rapid intestinal transit with intestinal hypersecretion can dilute the pertechnate and wash it out faster than it can be caught on image. In addition, active bleeding can result in reactive vasoconstriction in the diverticulum that can limit the delivery of the tracer. Swaniker et al. described 50% false negative results in patients with haemoglobin levels lower than 11 g/dl (24). On the other hand, Suh et al. reported that both sensitivity and specificity rise up to 100% once rectal bleeding in association with anaemia is considered, indicating that the bleeding should be abundant enough (25). Furthermore, false negative results have also been described due to an area of gastric mucosa smaller than 2 cm2, which may be too small to be visualised on the scan (11). However, other authors claim that the quality of nuclear medicine has improved over the last years and this should currently not be an issue (26). Finally, mucosal ischemia or necrosis may also result in a false negative scan. False positive results have been described due to intestinal duplications, obstructions, intussusception, ectopic kidney, vascular tumours, arteriovenous malformations and inflammatory foci (11). In cases of obstruction or inflammation caused by MD, the presence of gastric mucosa is less common, therefore test results for a Meckel scan will rarely be positive and its use in this context is not recommended (3).

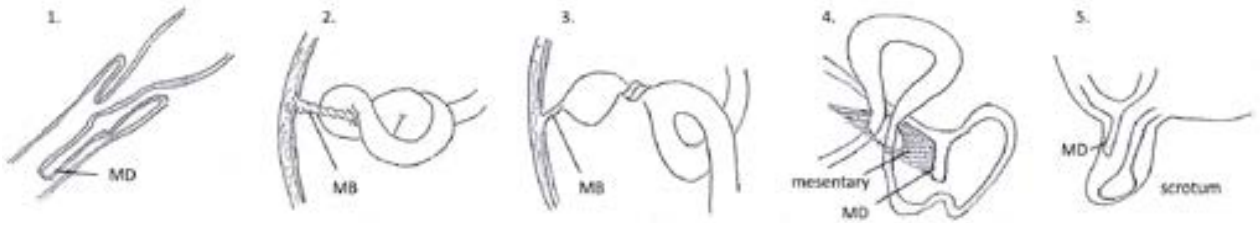
In order to increase the diagnostic value of the Meckel scan, premedication with certain drugs has been introduced. Histamine-2 (H2)-receptor

Table 2. Overview of clinical presentation forms and proposals for diagnostic investigations.

	Obstruction		Intestinal hemorrhage	Inflammation	
	Non intussusception	Intussusception		Diverticulitis	Perforation
Symptoms					
Abdominal pain	+	Intermittent	No pain	Appendicitis-like	+
Nausea/vomiting	+	+		+	+
Rectal bleeding		+	Massive, dark coloured		
Anaemia			+		
Fever				+	
Diagnostic investigations					
Radiography	+	Unable to reduce on contrast enema			+
Ultrasound		+		Actively rule out appendicitis	
CT scan	+			+	+
Meckel scan			+		
Laparoscopy	+	+	+	+	+

Non intussusception forms of obstruction: volvulus, strangulation due to torsion of fibrous bands, kinking, knotting and internal intestinal hernias

Figure 2 : *Types of obstruction caused by MD.*
1. Intussusception, 2. Volvulus around Meckel's band, 3. Strangulation by torsion of Meckel's band, 4. Internal hernia through the mesentery of MD, 5. Littre hernia. MD = Meckel's diverticulum, MB = Meckel's band.
Adapted from Netterimages: Meckel's Diverticulum: Complications of Meckel Diverticulum and Vitelline Duct Remnants.



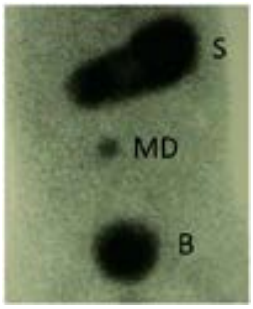
antagonists (e.g. ranitidine) inhibit the release of pertechnate into the bowel, concentrating the signal in the MD. Tseng et al. saw a 11% higher yield of positive Meckel scans in children with 3-day oral H2-blocker use prior to examination than those without (75% vs 64%) (15). Irvine et al. could increase the sensitivity and specificity to 100% using ranitidine for 24 hours prior to the examination (26). No studies with proton pump inhibitors in this context have been found. Other forms of premedication include pentagastrin that enhances mucosal uptake of pertechnate into the gastric mucosa, and glucagon that slows down small bowel motility (27). A repeated Meckel scan is recommended if the initial scan is negative or equivocal, but clinical suspicion for MD is still high or patient preparation was inappropriate.

There are other examinations less commonly used for diagnosing a bleeding MD. Tseng et al. was able to detect the bleeding source thanks to a red blood cell scan in four out of five patients with a negative Meckel scan (15). Magnetic resonance enterography has been successful in diagnosing a bleeding MD in 4/25 children with obscure intestinal bleeding (28). Double balloon enteroscopy can be a reliable diagnostic tool for a bleeding MD in children with a negative Meckel scan, as described in a small case series in China (29). Access to this technique, however, remains an important limitation. Finally, video capsule endoscopy (VCE) has been shown to be useful in the diagnosis of a bleeding MD. In a recent study by Wu et al., 15/37 children with intestinal bleeding from MD were positive on VCE, of which only 12 by Meckel scan (30). Despite limited evidence, VCE can potentially be of added value in the diagnosis of a bleeding MD. Shortcomings of VCE are the limitation of the patient's age range, potential risks of delayed passage and obstruction requiring surgical removal of the device (31). In Belgium, VCE is only reimbursed when there is a need for blood transfusion.

Table 3. Proposed recommendations for practical use.

Recommendation
In case of acute abdominal pain, · offer radiography to rule out obstruction or perforation, · offer ultrasound to rule out appendicitis, · consider CT scan if former investigations are insufficient or consider laparoscopic exploration.
When intussusception is suspected, offer barium reduction for resolution and be aware of relapse.
In case of failed reduction with barium enema or relapse of intussusception, offer laparoscopic exploration.
In case of rectal bleeding, · consider upper and/or lower endoscopy. · offer a Meckel scan with premedication. · offer a second Meckel scan when the scan is negative and clinical suspicion remains.

Figure 3 : *Meckel scan. The Meckel scan shows physiological enhancement in the stomach (S) and bladder (B). In the right upper quadrant the Meckel's diverticulum (MD) appears. S = stomach, MD = Meckel's diverticulum, B = bladder.*



Rectal bleeding is not specific for a bleeding MD. Obviously, upper and/or lower endoscopy should be considered during diagnostic workup for intestinal haemorrhage to investigate intestinal polyps, inflammatory bowel disease or other pathology.

Diagnosing the inflamed MD

Meckel diverticulitis can be seen on a CT scan as a cystic mass with surrounding inflammation. On ultrasound, a thick wall mass with a "gut signature" is visible, but can easily be mistaken for an appendicitis (22). The appendix should be actively sought on imaging to rule out appendicitis. In case of perforation, free intraperitoneal air can be seen on plain radiograph or CT scan. Radiation exposure remains an important concern in children, so limited use should be promoted.

Management

Once MD is suspected or diagnosed in a symptomatic patient, surgical excision is necessary. This can be performed by diverticulectomy, wedge-shaped excision or segmental bowel resection followed by end-to-end anastomosis. It is key to make sure there is no remaining ectopic mucosa left that can further harm the intestine, and sites of ulceration or gangrene tissue should be excised completely. Commonly, a long MD with a length-width ratio of more than 2 cm will have ectopic tissue at the tip or body and not at the base. In this case a simple diverticulectomy is proposed. In contrast, when the diverticulum is short or broad-based, gastric mucosa is likely to be located at the base of the MD and a broader approach is necessary (11,14). Other authors have proven that a diverticulectomy-only approach is sufficient and safe regardless of the size of the diverticulum (32,33). In cases of a bleeding MD, the bleeding site is typically adjacent to the ileal wall, and a wedge-shaped excision is often preferred (3). However, some studies showed that by performing a simple diverticulectomy, gastric heterotopia is completely resected and there is no increased risk of postoperative bleeding, a lower overall complication rate, and shorter operative times and hospital stays (34). Wedge or segmental resection is the preferred treatment of choice in children presenting with obstruction or diverticulitis with inflamed or perforated base.

The procedure can be done laparoscopically when diagnosis is certain or following exploratory diagnostic evaluation. This minimally invasive surgical technique is safe and associated with short hospital stays with minimal complication rates (8). In order to avoid laparotomy, Menezes et al. promotes an alternative method: the authors performed resection of MD in five patients laparoscopically but brought the diverticulum out of the abdomen via the umbilical port to perform resection-anastomosis without doing a laparotomy (11).

It remains controversial whether all incidentally diagnosed MDs should be resected. Some authors suggest resection of all MDs because of potential life-long risk for complication and low risk associated with laparoscopic resection. Although postresection complications are rare, intestinal adhesion obstruction and wound infections can occur. The main argument not to resect is the low risk of MD becoming symptomatic. Based on a systematic review, 758 resections are required to prevent one death from MD (2). Other authors recommend resection only when there is suspected ectopic gastric mucosa or adhesive bands. Slivova et al. showed that the presence of gastric heterotopia was associated with the width of the diverticulum. They recommend to resect an asymptomatic MD when the width of the diverticulum is more than 1,5 cm (34).

Pathological findings

MD is called a true diverticulum since it comprises all four layers of the intestinal wall: mucosa, submucosa, muscle and peritoneal serosa as the surface. In about half of the cases (28-84%), it contains ectopic or heterotopic tissue that can lead to complications (3-6,8-15). Gastric mucosa is the most common type of ectopic tissue (in 75% of cases) followed by pancreatic mucosa or a combination of gastric and pancreatic mucosa. Only 1 paediatric patient with colonic tissue has been reported (5).

MDs with ectopic tissue are more likely to become symptomatic, 50-60% in comparison to 4,6% in the overall MD population (4). Acid secretion from gastric mucosa or pancreatic enzymes from pancreatic tissue can cause bleeding and ulceration (35). In contrast, MDs complicated by obstruction or inflammation less frequently contain ectopic tissue. Ectopic pancreas tissue, however, has been shown to be a lead point for intussusception (3).

In very rare occasions, tumours can arise in children with MD, such as adenocarcinoma, carcinoid tumors, desmoplastic small round cell tumors, and benign mesenchymal tumors (lipoma, hemangioma, and hamartoma) (36).

Conclusion

MD is an infrequent congenital anomaly that generally remains asymptomatic during lifetime. However, it can lead to serious life-threatening complications, such as intestinal obstruction (mainly caused by intussusception), haemorrhage, and inflammation. Although complications from MD are possible at any age, they occur more often in children. Several diagnostic imaging studies are available but will only indirectly indicate the presence of MD by detecting its complications, and generally have a low accuracy to reveal the MD itself. During diagnostic workup for abdominal symptoms, MD should be considered for prompt diagnosis and treatment. Important characteristics specific for MD are 1) dark, massive rectal bleeding causing anaemia and often the need for transfusion, 2) the inability to reduce an intussusception on contrast enema or subsequent relapse, 3) clinical suspicion for appendicitis which is ruled out on ultrasound. In case of rectal bleeding and/or anaemia, a Meckel scan can support the diagnosis of a bleeding MD with a high sensitivity and specificity, and should be repeated when the result is negative and suspicion remains. Finally, laparoscopy can be a diagnostic modality of choice in children suspected to have MD. As for the treatment, a laparoscopic diverticulectomy-only approach has been found to be sufficient and safe in most cases.

Conflict of interest

The authors have no conflict of interest to declare.

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