

# Unusual presentation of obscure Meckel diverticulum treated with robot-assisted diverticulectomy

# A case report

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### Abstract

**Introduction:** Meckel diverticulum (MD) is the most common congenital abnormality of gastrointestinal tract. Tough believed to occur in 2% of population, most of them remain veiled because majority are clinically asymptomatic and remain obscure in radiological examination.

**Clinical Findings and Diagnosis:** A 26-year-old male with episodic black colored stool since last 10 years. Tough symptomatic, diagnosis of pathological lesion, and the bleeding site could not be established with any of the sophisticated diagnostic technique. After 10 years, it was finally diagnosed as MD with careful observation of bowel loops on computed tomography enterography (CTE) where remnant of vitelline vessel and hyper-enhancing nodule are seen along the wall of diverticular loop.

**Interventions and outcomes:** The patient underwent robot assisted laparoscopic surgery with excision of diverticular loop. To the best of our knowledge, this robot-assistant Meckel diverculectomy is probably the first reported surgical procedure in PubMed. Follow-up for 3 month showed no complication or recurrence.

**Conclusion:** Every case is unique and we must be aware and remain alert in tracing the possible morphological variation of the case. Here, we present one unique but rare feature of MD, which helped us in making diagnosis.

**Abbreviations:** CTE = computed tomography enterography, DBE = double balloon enteroscopy, GI = gastrointestinal, MD = Meckel diverticulum, VID = vitello-intestinal duct, WCE = wireless capsule endoscopy.

Keywords: CT enterography, Meckel diverticulum, omphalomesenteric tract, vitelline artery, vitello-intestinal duct

# 1. Introduction

Meckel diverticulum (MD) is a congenital anomaly of the gastrointestinal (GI) tract. It occurs due to failure of complete

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Vedio1. Meckel's diverticulum treated with robot-assisted diverculectomy.

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obliteration of vitello-intestinal duct (VID).<sup>[1]</sup> Tough believed to occur in 2% of population, most of them remain veiled unless any complication occurs.<sup>[2]</sup> GI bleeding is the most common complication seen with the MD. Most of them remain obscure with conventional radiological test, and it is also hard to trace out with upper GI endoscopy and colonoscopy. Angiography could be positive in case of active bleeding, and enteroscopy could be other sensitive method. In uncomplicated cases, nuclear imaging is thought to be the most sensitive test to diagnose MD because majority of cases are positive for gastric type mucosa within the diverticulum.<sup>[3]</sup> Here, we present one unique case of MD treated with robot-assisted diverticulectomy. To the best of our knowledge, this is the first reported case of MD excised using robot-assisted diverculectomy in Pubmed. We also discuss other possible techniques to help make diagnosis and share some images to strengthen the understanding of MD.

## 2. Case report

A 26-year-old male was admitted to our hospital (The First Affiliated Hospital of Sun Yat-Sen University) with complain of black tarry stool for 1 month. He complained of past history of similar symptoms since last 10 years and was episodic. The 1st time he presented with black stool was on 2006. He then went to local hospital, where upper GI endoscopy was done, but no significant positive signs for black stool were found. The symptoms were relieved with some medicine, which the patient does not remember and was discharged from hospital. Similar symptoms recurred in 2007 and again in 2010. Every time, just the symptomatic treatment was done and then the patient was discharged home.

On August 2011, capsule endoscopy was performed to rule out the intestinal cause of GI bleeding. It showed diffused hyperemic mucosa in terminal ileum, but no obvious erosion/ulceration or bleeding site was seen. Inflammation of terminal ileum was suggested, but the actual diagnosis was not made. On 2013, the patient again presented with black stool. Upper GI endoscopy was done again and it showed nonatrophic gastritis. Since no ulcer or any source of bleeding detected, enteroscopy was planned. On double balloon enteroscopy (DBE), large amount of dark red fluid and blood clots like materials were seen in small intestine. Concerning the severity of the bleeding, emergency digital subtraction angiography was done, where no active extravasation of the contrast medium was seen in bowel loop. As before, just the symptomatic treatment was prescribed and discharged home. On 2015, the patient again presented with black stool. Colonoscopy and DBE was done and all the bowel mucosa was visualized, but no abnormality detected except small localized hyperemia in the ileum. Computed tomography enterography (CTE) scan was also performed and no significant positive finding detected. Again just the symptomatic treatment was prescribed and then discharged home.

On January 2016, the patient presented with similar episode again, and then he referred to our hospital for further evaluation. At the time of admission he did not had active GI bleeding or black stool and no other associated symptoms. Physical examination was normal. Laboratory examinations showed all parameters within normal limits. Stool test for RBC and occult blood was also negative. On CTE scan with neutral oral contrast agent, loop of normal appearing bowel is seen. In the arterial phase, 1 normal appearing bowel (near ileum) showed high attenuated blood vessel along a small segment of a bowel wall (Fig. 1). Also the wall in some region is seen thicker than the rest of the bowel wall and a hyper-attenuated nodule is seen at the tip of the blind loop (Fig. 2). On Careful observation the loop is seen extending out from ileum and ends as a blind loop. He was further advised to do positron emission tomography–computed tomography (PET/CT) and barium meal. On barium study, a small outpouching from ileum is seen (Fig. 3). PET/CT using technetium-99m pertechnetate showed high uptake in stomach and physiological excretion in urinary bladder, but the diverticular lesion did not showed any uptake (Fig. 4). 3-D reconstruction image showed a diverticular loop originating from ileum, and a prominent branch of blood vessel from superior mesenteric artery is seen supplying the diverticulum (Fig. 5).

The patient underwent robot-assisted abdominal laparotomy (Video 1, http://links.lww.com/MD/B361). A 6 cm-long diverticulum was seen at the antimesenteric border of ileum. Small blood vessel surrounded by fibrous band was seen along the outer wall of the diverticulum, it was originated from mesentery and extended up to the tip of diverticulum. The base of the diverticulum was 2 cm wide and was slightly edematous. The tip of the diverticulum was hard but no attachment with surrounding tissues or abdominal wall seen. The diverticulum was excised at the base by the Endo-GIA stapler and sutured strongly. Frozen section during the surgery showed no mitosis or atypical cell. On postsurgical histopathological examination, fundus type gastric glands and pancreatic tissue were seen in submucosal, muscular, and serosal layers (Fig. 6). It was diagnosed as MD with ectopia of gastric mucosa and pancreatic tissue. The postoperative course of the patient was uneventful and the patient was discharged 7 days after the surgery. No any complication or recurrence was noted during 3 months' followup. This study was approved by the First Affiliated Hospital of



Figure 1. Axial view of abdomen showing blind loop of diverticulum (arrow head) at right iliac region. The wall showed avid enhancement in compare to other bowel. A prominent blood vessel (arrow) is seen along the wall of diverticular loop.



Figure 2. Sagittal view of abdomen showing blind loop of diverticulum (arrow head) originated from ileum. A hyper attenuated nodule (arrow) is seen at the tip of the diverticular loop.



Figure 3. Barium study showing small intestine, cecum, and colon. Outpouching of a blind loop is seen (arrow head) originating from terminal ileum (I).

Sun Yat-Sen University Institutional Review Board. Written consent for this case report was obtained from the patient.

#### 3. Discussion

MD is the remnant of the prenatal yolk stalk (VID). Embryologically, the omphalomesenteric tract contains 3 structures: the vitelline duct, vein, and 2 arteries. VID involutes during 5 to 7 weeks of intrauterine life so do the vitelline arteries. Failure of complete obliteration of VID can result in remnants, called MD. Among the right and left vitelline arteries, the left involutes whereas the right persist as superior mesenteric artery and it terminally supplies the diverticulum.<sup>[4]</sup> Anomalies of the vitelline circulation are less common, the remnant vitelline artery have been reported in 8% to 15% of cases of MD.<sup>[5]</sup> Rarely, remnant of vitelline artery in absence of MD is also possible.<sup>[1]</sup> The remnants manifest as peritoneum covered fibrous bands which extent from ileal branch of superior mesenteric artery either to a MD (mesodiverticular band) or to the anterior abdominal wall at the umbilicus. These bands may be completely patent, segmentally patent, or not patent.<sup>[6]</sup> Our case describes a rare case of MD with mesodiverticular band and remnant of patent vitelline artery within the band.

It is located in the distal ileum and is usually 60 to 100 cm away from the ileocecal valve. Embryologically, vitelline duct is made up of pluripotent cell lining, therefore may harbor multiple type of other tissue within the true diverticular loop. The commonst ectopic tissue found within MD is gastric mucosa, less commonly ectopic pancreatic or duodenal mucosa is present.<sup>[7]</sup> In our case, both the fundus type gastric gland and pancreatic tissue is seen in the diverticular loop. The reported incidence of ectopic mucosa in MD varies; a higher incidence is reported in patients who present



Figure 4. Positron emission tomography-computed tomography (PET/CT) dynamic imaging demonstrating persistent focus of technetium-99 m pertechnetate (radiotracer) uptake in stomach and its high activity in bladder following renal excretion. No uptake of tracer is seen in any other bowel.



Figure 5. 3-D reconstruction image showing a diverticular loop (black arrow head) originating from ileum (I) and prominent branch of blood vessel (white arrow head) from superior mesenteric artery (arrow) is seen supplying the diverticulum.

with GI hemorrhage (55%–100%) and in pediatric patients (61%).<sup>[3,8]</sup>

Prevalence of MD is believed to be approximately 2% of general population, with 3 to 5 times higher prevalence in male than female.<sup>[9]</sup> Among them, majority remained asymptomatic, only 4% cases are symptomatic and are usually seen secondary to development of complications.<sup>[10]</sup> Gastrointestinal bleeding (28%) is the most common complication of MD, followed by intussusception (13%), obstruction (11%), and perforation (11%).<sup>[11]</sup> Other complication such as strangulation, diverticulitis, volvulus, internal hernia, and neoplasm is also reported.<sup>[11]</sup> Symptoms are often present among children below 2 years of age, the risk will decrease with increasing age.<sup>[12,13]</sup> But our case was a 26 year male and he had a GI bleeding since age of 16.

Hemorrhage is the most common complication of MD and is believed to occur due to embryonic remnant of gastric and pancreatic mucosa in the diverticulum. Secretion of acid from gastric remnant and alkaline juice from pancreatic remnant leads to ulceration in the adjacent ileal mucosa.<sup>[14]</sup> In rare instant, mechanical stimulation could also be the cause of erosion and ulceration. Tumors in MD may cause bleeding. Benign tumors (leiomyoma, lipoma, vascular, and neurovascular hamartoma) or malignant tumors (carcinoids, mesenchymal tumors, adenocarcinoma, small round cell tumor) all can complicate with MD bleeding. Other causes of melena must also be ruled out.



Figure 6. Histopathological specimen showing heterotopic pancreas tissue (A) and heterotopic gastric glands (C) in submucosal, muscular, and serosal layers. High-power field showing prominent pancreatic tissue (B) and gastric gland (D).

Often, the 1st approach is to use endoscopy to look for obvious signs of upper GI bleeding. For lower GI bleeding, colonoscopy is often the 1st choice. Our case underwent upper GI endoscopy for many times and the colonoscopy was done once, but the cause of bleeding was not found. Targeted red blood cell scan and mesenteric angiogram can also be used to detect the source of bleeding in case of normal endoscopy and colonoscopy findings. With mesenteric angiography, active extravasation of contrast medium into the bowel can only reliably be seen when blood loss exceeds 0.5 mL/minute. In contrast imaging using technetium-99m labeled red blood cells can identify hemorrhage at rates as low as 0.1 mL/minute. In our case, digital subtraction angiography was done once when abundant blood and blood clot like material is seen in enteroscopy, but the site of active bleeding could not be seen on angiography.

DBE and wireless capsule endoscopy (WCE) is an alternative to angiography or scintigraphy in the investigation of obscure GI bleeding. Although the sensitivity of WCE in detecting an MD is not well established, there are several reports of WCE successfully identifying an MD as a source of GI hemorrhage.<sup>[15]</sup> Some authors claiming higher sensitivity and specificity of WCE for the diagnosis of MD-related obscure GI bleeding,<sup>[16]</sup> whereas other claim DBE and single-balloon enteroscopy are excellent. In our case, WCE was done once in 2011 and DBE was done on 2013 but neither of the modality helped to find the source of GI bleeding and the presence of MD. It could be for there was no active bleeding at the time of examination.

Radiology plays a variable role in the diagnosis of MD. Thought different imaging modality available, radiological diagnosis of an MD is still a challenge. Plain radiographs are not usually helpful in making the diagnosis of an MD.<sup>[17]</sup> They may be normal or show nonspecific signs in case of complications. Barium study can be helpful. The characteristic appearance of an MD is a saccular structure attached to the antimesenteric border of distal ileum. But it is never the 1st choice in the case of complicated cases with GI bleeding and acute abdominal pain and overall sensitivity of contrast studies in detecting an MD varies.

Ultrasound offers a noninvasive and radiation free procedure with dynamic information. Several sonographic features suggestive of MD have been described, but none of them are specific. The sonographic appearances of MD depend on the complications then the primary lesion. Thus, ultrasound is not much helpful in making diagnosis of an MD, but it can guide surgeons, whether surgical or conservative management to be employed.

CT appearances of MD usually resemble a normal bowel loop.<sup>[18]</sup> Complications will determine the CT appearance. Inflammation appears as a thick-walled loop of bowel with associated inflammatory change in the surrounding mesentery, but normal appendix.<sup>[19]</sup> Hemorrhage from an MD using CT angiography has been reported while conventional angiography was negative.<sup>[20]</sup> If a lesion is suspected in small bowel and conventional CT showed no abnormality, CT enteroclysis/ enterography (CTE) could be diagnostic. CTE has a combine advantages of assessing the mucosa and lumen along with the assessment of extra-luminal pathology. This techniques have been used to successfully identify an MD.<sup>[21]</sup> Here, we diagnosed the case on CTE, remnant of vitelline duct was seen along with prominent blood vessel and irregularly thickened bowel wall in the arterial phase. The artery runs along the wall of the diverticular loop and ends with the closed loop of Meckel diverticular wall. We also find avid enhancement in the diverticular wall and it represents hypervascular mass. The hypervascular mass in MD could be pancreatic tissue, neuroendocrine tumor, or GIST. Pancreatic tissue is found in the postsurgical histopathological report of our case.

Nuclear medicine studies (Tc-99m pertechnetate scan, known as Meckel scan) remain one of the most specific diagnostic tests in MD. Ectopic gastric mucosa within an MD is seen with scintigraphy due to the uptake of technetium-99m pertechnetate by mucus secretory cells.<sup>[22]</sup> The sensitivity of scintigraphy could be improved by the use of H2 antagonists, pentagastrin, or glucagon. The common cause of negative Meckel scan could be absence of gastric mucosa or could be due to absence of specific cell within the abundant gastric mucosa that uptakes pertechnetate, thus showing no appreciable uptake on scintigraphy.<sup>[23]</sup> In our case, ectopic gastric mucosa is shown to be present on histological report but the scintigraphy showed negative uptake. It could be due to absence of the specific cell that has high uptake of pertechnetate.

Compare to open surgery, laparoscopic surgery is believed to be safer, feasible, with improved outcome, shorter hospital stay, and reduce cost.<sup>[24]</sup> Laparoscopically, excision of the MD could be treated by using endostaplers, or extracorporeal bowel segment resection. Segmental bowel resection is preferred if the base of diverticulum is broad ( $\geq 2$  cm), inflamed, or in case of bleeding ulcer. Furthermore, surgeon must be extra cautious to handle the special case like ours where there is patent vitelline artery along the wall of the bowel. Hemorrhagic complication could be life threating if the diverticulum with patent vitelline artery is resection inadvertently. Despite all the risk factors, our case underwent robot-assisted diverculectomy using Endo-GIA stapler for the 1st time in our hospital, and it was successfully conduction with no complication. Robot-assisted diverculectomy had been practiced to treat bladder, esophageal, and calyceal diverticulum, but probably this is 1st case reported to treat MD with robot-assisted diverculectomy.

Compare to plain film, ultrasound, and noncontrast CT, contrast studies of the small bowel can have higher sensitivity in diagnosing diverticulum; however, this is never a first choice in complicated abdominal cases. Technetium-99m pertechnetate scintigraphy is believed to be most sensitive test in the diagnosis of MD. Mesenteric angiography performed for GI bleeding could show negative result in case of inactive bleeding. WCE and DBE is a newer technique and can be useful in selected patients. Thus, sensitivity and specificity of individual test varies according to the clinical and morphological variations. Therefore, it is advisable to do multiple tests. Careful observation of every possible morphological variations and radiological diagnostic sign is mandatary to make diagnosis with high accuracy. Newer technique like robot-assisted diverculectomy could be safe and feasible treatment modality despite some risk factors.

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