J Ped Surg Case Reports 3 (2015) 185–189

Contents lists available at ScienceDirect

Journal of Pediatric Surgery CASE REPORTS

journal homepage: www.jpscasereports.com

An unusual case in which a perforated Meckel's diverticulum became trapped in a pericecal hernia: A rare complication of Meckel's diverticulum

Satohiko Yanagisawa*, Yasuhide Morikawa, Mototoshi Kato

Department of Pediatric Surgery, International University of Health and Welfare Hospital, 537-3, Iguchi, Nasushiobara, Tochigi 329-2763, Japan

ARTICLE INFO

Article history:
Received 30 November 2014
Received in revised form
25 February 2015
Accepted 1 March 2015

Key words:
Meckel's diverticulum
Complication
Internal hernia
Pericecal hernia
Child

ABSTRACT

An 11-year-old boy had previously been diagnosed with repeated Meckel's diverticulitis at another hospital. Emergency laparoscopy was performed under general anesthesia, and an inflammatory mass was seen in the ileocecal region of the mesentery. However, no Meckel's diverticulum (MD) was observed, and so the patient was diagnosed with lymphadenitis. Three days after the operation, he developed anemia and gastrointestinal bleeding of unknown origin. Thus, he was transferred to our hospital for further investigation and to have his gastrointestinal bleeding treated. Based on imaging scans obtained at the previous hospital, a paraduodenal hernia was suspected, but no paraduodenal hernia was detected during emergency surgery, despite the fact that the full length of the normal small intestine could be traced. However, an inflammatory mass was observed, and the ileum appeared to be incarcerated in a pericecal hernia. We could not identify which portion of the intestine had become entrapped or reduce the hernia due to adhesion. The inflammatory mass was removed by ileocecal resection, and a pathological examination revealed that the entrapped portion of the intestine was an MD that had branched off from the small intestine immediately proximal to the ileocecal valve. The MD had perforated in the hernia sac, which had caused the patient's bleeding.

Meckel's diverticulum (MD) is a common cause of abdominal pain, obstruction, and gastrointestinal bleeding among children. The incidence of MD is approximately 2–4% among the general population [1–5], and approximately 4–6% of patients with MD develop complications that require emergency surgery. These complications include obstruction, gastrointestinal bleeding, diverticulitis, perforation, and tumors. Internal hernias are an exceedingly rare complication of MD. We report an unusual case in which an MD had become herniated into a pericecal hernia and perforated. The imaging findings of this case were difficult to distinguish from those of paraduodenal hernia.

1. Case report

An 11-year-old boy had suffered from intermittent abdominal pain for 10 days. As his symptoms had got worse, he was admitted to a local hospital. He was diagnosed with Meckel's diverticulitis based on the findings of computed tomography (CT) (Fig. 1) and was conservatively treated with antibiotics. His symptoms disappeared after 7 days of treatment. However, he experienced severe abdominal pain on the night that oral intake was re-started. The patient's symptoms were ameliorated by a few days of fasting, but the abdominal pain recurred immediately after he consumed a meal. Repeated Meckel's diverticulitis was suspected based on the findings of a second CT scan (Fig. 2), and emergent laparoscopy was performed on day 16. The small intestine could be traced from the ileocecal valve to the ligament of Treitz, and no MD was observed. Thus, the patient was diagnosed with lymphadenitis, as an inflammatory mass had been detected in the ileocecal region of the mesentery. On the third postoperative day, a copious bloody fecal discharge was observed, which continued for 3 days. Anemia (hemoglobin: 7.5 g/dL) was detected by a laboratory test. Upper gastrointestinal endoscopy and a small bowel follow-through examination were performed, but the cause of the patient's bleeding could not be identified.

Thus, the patient was transferred to our hospital for further investigation and to have his gastrointestinal bleeding treated. On admission, he could not walk due to the effects of protracted bed

* Corresponding author. Tel.: +81 0287 37 2221; fax: +81 0287 37 5315.
E-mail address: s.yanagisawa@jichi.ac.jp (S. Yanagisawa).

http://dx.doi.org/10.1016/j.epsc.2015.03.003

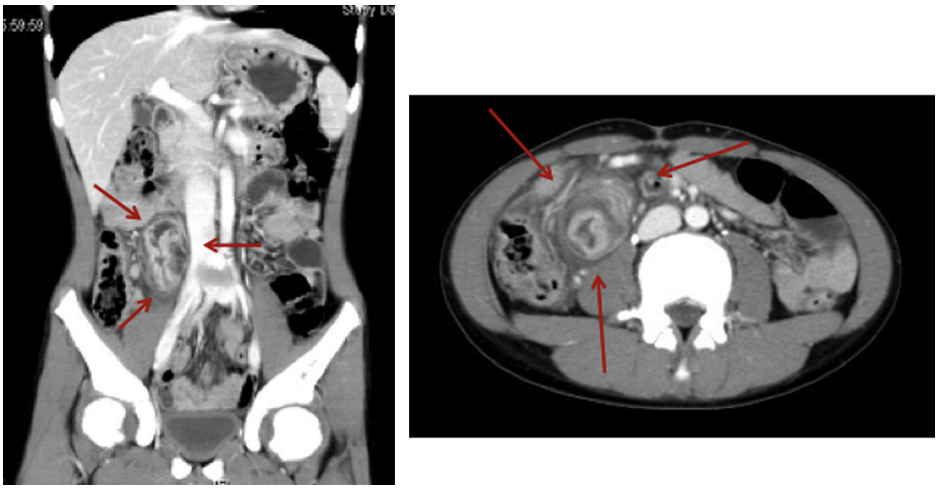

Fig. 1. CT scan obtained on admission. A dilated intestinal structure (diameter: 6.5 × 4.2 cm) containing an enhanced layer was observed dorsal to the mesentery in the right lower quadrant (red arrow).

rest and long-term fasting. We did not observe any abdominal tenderness or melena. We reviewed the imaging scans obtained at the previous hospital. The CT scan performed at admission led us to suspect diverticulitis or intussusception of an MD (Fig. 1). Furthermore, the CT scan performed after the patient had suffered severe abdominal pain revealed that an intestinal structure in the right lower quadrant had spread to the cranial quadrant and had pushed the superior mesenteric artery aside (Fig. 2). Based on the patient's CT findings, we suspected a paraduodenal hernia. However, a small bowel follow-through examination performed after upper gastrointestinal endoscopy did not detect any findings that were indicative of obstruction of the small intestine or MD. We suspected that the inflammatory mass that had been detected in the ileocecal region during the laparoscopic examination might have been the intestinal structure observed on the abovementioned CT scan. As no MD was detected during the laparoscopic examination, we considered that it was possible that the intestine had become incarcerated in a paraduodenal hernia, which had then reduced spontaneously. We assumed that the patient only experienced abdominal pain when the intestine was incarcerated.

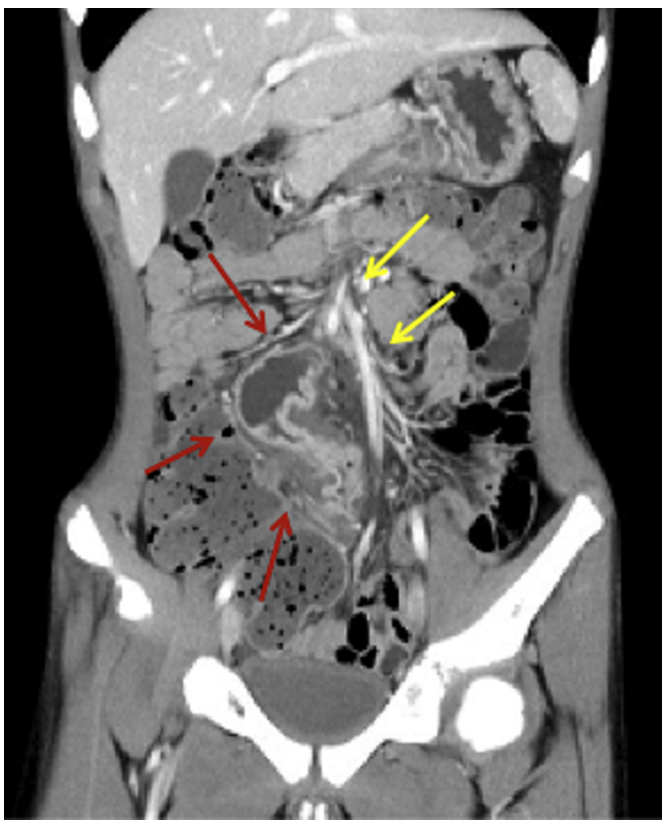

Fig. 2. CT scan obtained after the patient experienced abdominal pain. CT showed that the intestinal structure in the right lower quadrant (from Fig. 1) had spread to the cranial quadrant and pushed the superior mesenteric artery aside (yellow arrow).

The day after the patient was transferred to our hospital, an emergent laparotomy was performed via an upper abdominal transverse incision, which revealed a normal small intestine from the ileocecal valve to the ligament of Treitz. No paraduodenal hernia was observed. However, an inflammatory mass was observed in the ileocecal region, and palpation confirmed that it was an entrapped section of the intestine (Fig. 3A). Unfortunately, we could not identify which portion of the intestine had become entrapped. The ascending colon and the right side of the transverse colon were mobilized by dissection, using a similar procedure to that used to treat right hemicolectomy. The entrapped part of the intestine appeared to be a section of the terminal ileum located in the mesentery, and it had been incarcerated within the inferior ileocecal fossa (Fig. 3B). The entrapped intestine could not be reduced because it had perforated and adhered tightly to the surrounding tissue. Therefore, we decided to resect the inflammatory mass by ileocecal resection.

A subsequent examination of the resected specimen demonstrated that the entrapped portion of the intestine was an MD (approximately 10 cm long), which had been perforated at its base (Fig. 4). The pathological findings of the resected specimen were typical of an MD, e.g., it exhibited a five-layered intestinal structure. Ectopic gastric mucosal tissue was also observed in the diverticulum, which had led to the development of a deep ulcer near to the base of the diverticulum and the subsequent perforation of the MD. The MD was located immediately proximal to the ileocecal valve. The MD was entrapped in a pericecal hernia, which is a rare type of internal hernia, and the entrapped section had perforated. Accordingly, the MD had also caused the patient's gastrointestinal bleeding. As only the MD was entrapped and had formed an inflammatory mass in the right lower quadrant, the normal

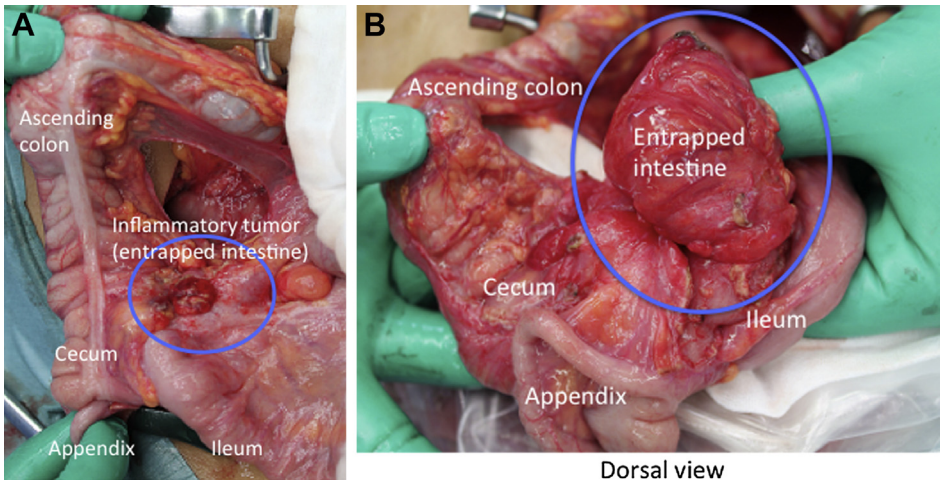

Fig. 3. Operative findings. A) An inflammatory mass was located between the ileocecal region and the mesentery (blue circle). B) After mobilizing the right colon, an inflammatory mass was observed on the dorsal side. The entrapped intestine had herniated into the inferior ileocecal recess.

intestine could be traced from the ileocecal valve to the ligament of Treitz.

The patient required total parenteral nutrition due to malnutrition caused by long-term fasting. On the fourth postoperative day, a dirty drain discharge led us to suspect a minor intestinal leak, although no additional contamination was observed. Therefore, oral intake was initiated on postoperative day 10, along with a rehabilitation program aimed at treating the patient's muscular atrophication. On postoperative day 21, the patient's nutritional status and his ability to perform activities of daily living had recovered to pre-admission levels, and we began to discuss a discharge schedule. However, strangulation of the intestine suddenly occurred, and laparotomy was performed. Adhesion between the intestine and mesentery around the anastomosis appeared to have caused the intestinal obstruction. The obstruction was reduced, and the patient's subsequent postoperative course was uneventful. He was discharged on postoperative day 49.

2. Discussion

MD is one of the most common congenital anomalies of the gastrointestinal tract, with an incidence of approximately 2–4%

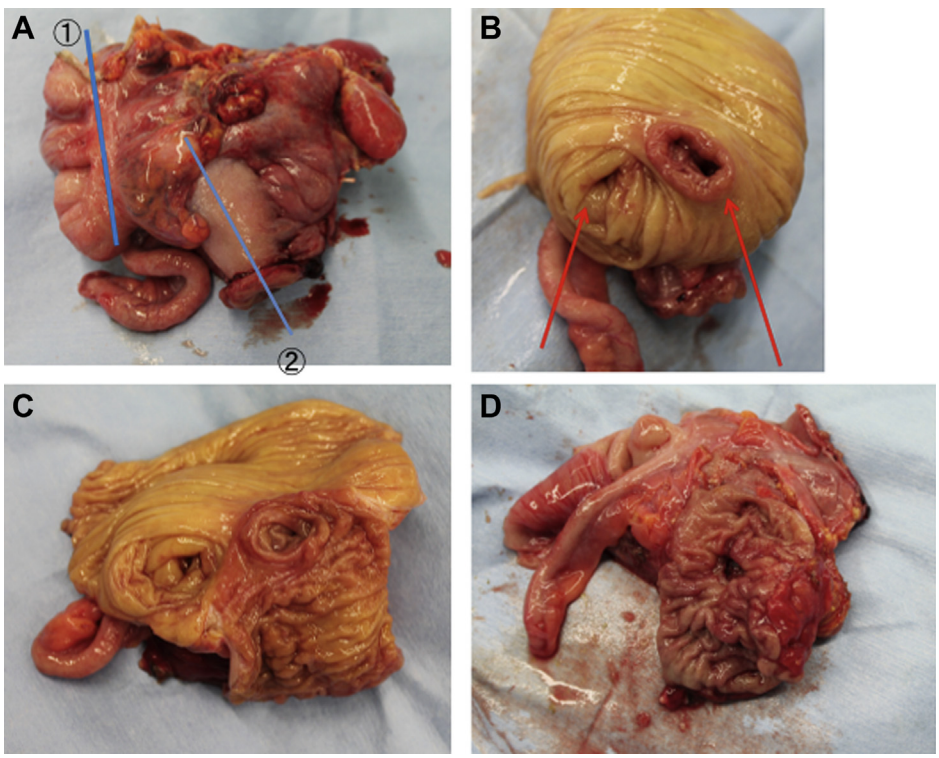

Fig. 4. The resected specimen. A) The resected specimen from the ileocecal lesion ① and ②: the resection lines. B) After cutting the cecal wall, two holes were visible in the ileocecal valve. One was the terminal ileum, and the other was the orifice of the MD, which branched off from the intestine immediately proximal to the ileocecal valve. C) After the wall of the terminal ileum had been resected, it was found that the orifice of the MD was located within 1 cm of the ileocecal valve. D) The 10-cm long MD seemed to correspond to the entrapped intestinal structure seen on CT. The MD had perforated near to its base.

among the general population [1–5]. Although MD are asymptomatic in most cases, complications develop in 4–6% of cases, which can include obstruction, gastrointestinal bleeding, diverticulitis, perforation, and tumors in adults. In children, bleeding is the most common complication (accounting for 55.5% of complications), followed by obstruction (14.2%), perforation (9.5%), diverticulitis (6.3%), and patent vitello-intestinal ducts (14.2%) [6]. Approximately 50–60% of pediatric patients who develop symptoms are <2-years-old, and such patients require emergency surgery due to the life-threatening nature of MD complications.

The present case involved an unusual complication of MD and included some rare characteristics. As a result, it was difficult to obtain a correct diagnosis and to identify which portion of the intestine had become entrapped.

The present case involved an MD that had become trapped in an internal hernia, which is exceedingly rare [7–10]. Internal hernias are defined as the protrusion of the bowel through a normal or abnormal opening within the boundaries of the peritoneal cavity, and they are detected in 0.2–0.9% of autopsies [10]. Pericecal hernias account for 10–15% of internal hernias and are the second most common type of internal hernia after paraduodenal hernias. Pericecal hernias are often classified into four subgroups according to which paracecal fossa they are located in; i.e., the superior ileocecal recess, inferior ileocecal recess, retrocecal recess, or paracolic sulci [9,10]. The present case involved the herniation of an MD into the inferior ileocecal recess, which is exceedingly rare (only one previous case has been reported [7]).

Littre hernias, which are defined as the protrusion of an MD through a potential abdominal opening, are rare. Littre hernias are reported to occur in 1–3% of patients with MD [11]. They can occur at inguinal (50%); femoral (20%), umbilical (20%); and other abdominal sites (10%), such as the foramen of Winslow [11]. MD that become trapped in internal hernias at rare abdominal sites are also referred to as Littre hernias.

In the present case, the MD was located in the terminal ileum, approximately 1 cm away from the ileocecal valve. Two holes; i.e., the ileum and the orifice of the MD, were visible in the resected specimen from the ileocecal valve (Fig. 4). Interestingly, MD are typically located 30–90 cm from the ileocecal valve, although the locations of MD vary considerably. In a previous study, the mean distance from the MD to the ileocecal valve was 50.6 cm (range, 5–150 cm), and the distance between the two structures increased with age (<2-years-old, 34.1 cm; 3–19-years-old, 45.9 cm; >21-years-old, 66.6 cm) [4]. Furthermore, 72% of MD are located within 46–91 cm of the ileocecal valve, and only 4% are located within 15 cm of the ileocecal valve [5]. Therefore, MD that are located in close proximity to the ileocecal valve are extremely rare.

In the present case, the MD appeared to be located on the mesenteric side of the intestine. MD that arose in rare mesenteric locations have been reported in 3 previous cases [12–14]. However, none of the previous case reports offered satisfactory explanations for the embryological development of such MD. Embryologically, an anti-mesenteric location is considered to be one of the characteristics of MD. Thus, the authors considered that the present MD did not actually arise at a mesenteric location.

To summarize the present case, it was considered that a 10 cm portion of an MD, which was located in close proximity to the ileocecal valve, had herniated into the inferior ileocecal recess from the anti-mesenteric border of the ileum. Then, a deep ulcer caused by the presence of ectopic gastric mucosal tissue had penetrated the MD, and local peritonitis had developed. The patient's clinical history and symptoms, e.g., severe abdominal pain that recurred after the resumption of oral intake and anemia caused by copious bloody stools, corresponded with the operative findings. Alternatively, the MD might have entered the retrocecal space because the cecum was not tightly fixed to the retroperitoneum. Then, after the MD had been perforated by the deep ulcer the mesentery and retroperitoneum adhered to the MD due to localized inflammation. As a result, the MD appeared to be an entrapped pericecal hernia and formed an inflammatory mass. Three broadly accepted criteria for identifying MD have been reported: (1) the diverticulum should be located on the anti-mesenteric border of the ileum, proximal to the ileocecal valve; (2) a separate blood supply should exist in the form of a mesenteriolum; and (3) the diverticulum should contain all the layers of the small intestine [5]. Two of the three criteria were satisfied in the present case, but we did not detect a separate blood supply for the MD, and the vitelline artery and mesodiverticular band could not be identified due to inflammation and adhesion.

As the complications of MD vary, it is difficult to diagnose complicated MD using conventional imaging techniques [15]. The imaging findings (particularly the CT findings) of complicated MD typically resemble those of paraduodenal hernias, and it can be difficult to distinguish the MD in such cases, as was found in the present case. Laparoscopy is recommended for diagnosing and treating complicated MD [15]. Although we agree with this recommendation, a laparoscopic examination did not result in a definitive diagnosis in our case. In addition, the patient's laparoscopic findings led us to doubt whether the inflammatory mass contained an entrapped portion of the intestine. One of the limitations of laparoscopic examinations is that internal hernias cannot be reduced due to adhesion. However, a hybrid treatment involving double-balloon enteroscopy and a surgical procedure has been reported as a new strategy for treating small intestinal lesions, including MD [16]. This technique might have provided important information and prevented us from questioning whether the intestine had actually been entrapped.

3. Conclusion

We have reported a rare case in which an MD became trapped within a pericecal hernia (a Littre hernia). When an internal hernia is suspected, a perforated MD must be considered.

Consent

Written informed consent for the publication of this case report and the accompanying images was obtained from the patient's next of kin. A copy of the consent form is available upon request.

Conflicts of interest

None of the authors has any conflicts of interest to disclose.

Sources of funding

We did not receive any funding for this study.

References

[1] Uppal K, Tubbs RS, Matusz P, Shaffer K, Loukas M. Meckel's diverticulum: a review. Clin Anat 2011;24:416–22.
[2] Matsagas MI, Fatouros M, Koulouras B, Giannoukas AD. Incidence, complications, and management of Meckel's diverticulum. Arch Surg 1995;130:143–6.
[3] Kusumoto H, Yoshida M, Takahashi I, Anai H, Maehara Y, Sugimachi K. Complications and diagnosis of Meckel's diverticulum in 776 patients. Am J Surg 1992;164:382–3.
[4] Yamaguchi M, Takeuchi S, Awazu S. Meckel's diverticulum. Investigation of 600 patients in Japanese literature. Am J Surg 1978;136:247–9.
[5] Jay 3rd GD, Margulis RR, McGray AB, Northrip RR. Meckel's diverticulum; a survey of one hundred and three cases. Arch Surg 1950;61:158–69.
[6] Menezes M, Tareen F, Saeed A, Khan N, Puri P. Symptomatic Meckel's diverticulum in children: a 16-year review. Pediatr Surg Int 2008;24:575–7.
[7] Aggarwal BK, Rajan S, Aggarwal A, Gothi R, Sharma R, Tandon V. CT diagnosis of Meckel diverticulum in a paracolic internal hernia. Abdom Imaging 2005; 30:56–9.

[8] Ko SH, Tioa MM, Huang FC, Hsieh CS, Huang CC, Ng SH, et al. Internal hernia associate with Meckel's diverticulum in 2 pediatric patients. Am J Emerg Med 2008;26:86–90.
[9] Molto Aguado M, González Valverde FM, Barreras Mateos JA, Vázquez Rojas JL. Small intestinal strangulation due to a primary intestinal paracecal hernia. Hernia 2007;11:457–8.
[10] Ghiassi S, Nguyen SQ, Divino CM, Byrn JC, Schlager A. Internal hernias: clinical findings, management, and outcome in 49 nonbariatric cases. J Gastrointest Surg 2007;11:291–5.
[11] Skandalakis PN, Zoras O, Skadalakis J, Mirilas P. Littre hernia: surgical anatomy, embryology, and technique of repair. Am Surg 2006;72:238–43.
[12] Walczak DA, Falek W, Zakarzawski J. An uncommon location of Meckel's diverticulum or small intestine duplication? Case report and literature review. Pol Przeal Chir 2011;83:457–60.
[13] Sarioglu-Buke A, Corduk N, Koltuksuz U, karrabul M, Savran B, Bagci S. An uncommon variant of Meckel's diverticulum. Can J Surg 2008;51: E46–7.
[14] Segal S, Albrecht DS, Belland KM, Elister EA. Rare mesenteric location of Meckel's diverticulum, a forgotten entity: a case study about USS Kitty Hawk. Am Surg 2004;70:985–8.
[15] Prasad TR, Chui CH, Singaporewalla FR, Ong CP, Low Y, Yap TL, et al. Meckel's diverticular complications in children: is laparoscopy the order of the day? Pediatr Surg Int 2007;23:141–7.
[16] Tsuji Y, Maeda K, Ono S, Tazuke Y, Yanagisawa S, Usui Y, et al. A novel diagnostic and treatment strategy for small intestinal lesions in children: hybrid treatment using transumbilical minimal incision surgery combined with double-balloon enteroscopy. Pediatr Surg Int 2013;29: 357–61.