Unusual presentation of a Meckel’s diverticulum: A case report

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ABSTRACT

INTRODUCTION: Meckel’s diverticulum (MD) is the most common congenital malformation of the gastrointestinal tract. Intestinal obstruction is the lead presenting symptom in the adult population due to multiple causes (intussusception, incarceration, adhesions, strictures and torsion). Our patient had a complicated MD with an unique combination of risk factors and findings.

PRESENTATION OF CASE: We report an unusual case of an 18-year-old patient presenting with acute small bowel obstruction for several days, who developed focal peritoneal signs on right lower quadrant. On laparotomy, findings included a necrotic giant MD and a small bowel volvulus around a fibrous band that attached MD to the umbilicus. Segmental enterectomy with primary anastomosis was performed.

DISCUSSION: Axial torsion and gangrene of MD is the rarest complication. Its pre-operative diagnosis remains elusive as it can be clinically indistinguishable from other intra-abdominal inflammatory conditions. The correct diagnosis of complicated MD before surgery is often difficult because this condition can mimic other acute abdominal pathologies. There are several risk factors that can point to an accurate and early diagnosis, especially when combined with the appropriate imaging techniques, such as computed tomography with oral and intravenous contrast.

CONCLUSION: This complication remains underdiagnosed, often with delayed surgical intervention and sub-optimal treatment that leads to significant morbidity and mortality.

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1. Introduction

Meckel’s diverticulum (MD) is the most common congenital malformation of the gastrointestinal tract due to incomplete obliteration of the proximal portion of the omphalomesenteric duct in the 7th week of gestation [1]. This congenital anomaly has often been referred to by the “rule of twos”—usually located 2 feet proximal to the ileocecal valve, present before the age of 2, is seen twice as commonly in men as in women, and is found in 2% of the population [2]. It is the only true diverticulum of the small intestine, containing all layers of the small bowel wall. MD is mostly clinically silent, particularly in the adult. Several risk factors for developing symptomatic MD have been identified: male sex, age younger than 50 years, the presence of a diverticulum with 2 cm or more in length, or those that contained heterotopic mucosa [3]. When two, three, or four of these criteria were met, the proportion of symptomatic MD increased to 25, 42, and 70%, respectively [4]. Robijn et al. also included the presence of a fibrous attachment to the abdominal wall as risk factor [5].

Intestinal obstruction of various types is the most common presenting symptom in the adult population. Cases of giant MD (>5 cm) are relatively rare and associated with more severe forms of complications, especially with obstruction [6,7]. Axial torsion and gangrene of MD is an extremely rare complication [8,9].

We present a case of a giant MD with a fibrous attachment to the abdominal wall and axial torsion in an adult male whose initial presentation was small bowel obstruction.

2. Presentation of case

An 18-year-old caucasian male presented to the emergency department with abdominal pain and distension, oral intolerance and bilious vomiting for 24 h. He referred peri-umbilical pain and nausea for 15 days that progressively worsened in the last 48 h. He described the pain as crampy that was relieved after vomiting. History taking revealed previous episodes of abdominal pain and bloating. He had no relevant medical history or previous surgery. Family history was unremarkable.

On physical examination, he was febrile (38.5°C) and hemodynamically stable (blood pressure 127/83 mmHg, heart rate 87 beats/min, SpO2 98%). The abdomen was distended and bowel...
bowel and the MD. We performed a segmental ileal resection with primary anastomosis. Histology revealed small bowel with necrotic lesions and gangrenous MD.

The recovery was complicated with superficial surgical site infection (SSI) that responded to drainage and antibiotics. The patient was discharged within 10 days. At 6-month follow-up the patient was well and remained asymptomatic.

This case was reported in accordance with the CARE guidelines (Table 1) [10].

3. Discussion

Documented incidences of symptomatic MD range from 4% to 16% in large series [3,4,11,12]. In a retrospective study with 1476 patients from Mayo Clinic, 16% of all patients with MD were symptomatic and diverticulum length greater than 2 cm was associated with symptoms, among other features [4].

Intestinal obstruction is the most common presentation in adults, representing 40% of symptomatic cases. The most common cause of obstruction is intussusception with MD being the leading point, or a mechanical volvulus of the small intestine around a persistent fibrous band that attaches the MD to the umbilicus [8]. Obstruction has been found to occur more frequently with a giant MD [6,8]. The volvulus can also be incomplete and recurrent, resulting in repeated episodes of intestinal sub-occlusion, as it happened with our case. Other causes of obstruction include incarceration of a diverticulum in an inguinal hernia (Littre’s hernia), inflammatory adhesions, diverticular strictures and tumor-containing MD [4].

Axial twisting of MD around its narrow base is a rare complication. In addition to this, gangrene of MD, secondary to axial torsion, is an extremely rare phenomenon [8,9]. The anatomical configuration, especially the diverticular length and base diameter, is an important predisposition factor [3,7–9]. An elongated variant with a narrowed base is far more likely to result in torsion [7], whereas short, large-base diverticula are subject to foreign body entrapment [13]. In our case, the diverticulum was 10 cm long and 2 cm wide, which may have predisposed it for torsion. Axial torsion occurred around its narrow base, resulting in decreased blood supply and gangrene.

Fewer than 10% of cases of complicated MD in adults are diagnosed preoperatively [13–15]. The correct diagnosis of complicated

sounds were augmented. There was tenderness to palpation on the lower quadrants, mainly in the midline, but without rebound tenderness or guarding. No abdominal scars or hernias were present. Rectal examination was negative.

Laboratory testing revealed leukocytosis with neutrophilia and elevated C-reactive protein.

Upright abdominal plain radiography (Fig. 1) showed multiple air-fluid levels. Contrast-enhanced computed tomography (CT) scan (Fig. 2) revealed wall thickening and air-fluid levels compatible with small bowel obstruction, without apparent mechanical cause. Inflammatory bowel disease was suggested as a cause.

Initially treated with conservative measures (intravenous fluids and nil per os), on the second day the patient’s condition deteriorated. He referred onset of right lower quadrant pain with focal peritoneal signs on physical exam. He was taken up for emergent laparotomy. Findings included a necrotic giant MD and a fibrous cord between the umbilicus and the tip of the diverticulum around which the bowel twisted. The giant gangrenous MD, measuring 10 cm in length and with a 2 cm base, was found 50 cm proximal to the ileocecal valve (Fig. 3). The band was lysed, unfolding the

Fig. 1. Upright plain abdominal radiography showing air-fluid levels in the small bowel (upper left quadrant).

Fig. 2. Abdominal CT scan with distended small bowel loops, wall edema and air-fluid levels.

Fig. 3. Necrotic Meckel’s diverticulum with the fibrous band attaching its fundus with the abdominal wall. Note the dilated proximal small bowel loops in contrast with the distal ones indicating that the diverticulum acted as a torsion point.
MD before surgery is often difficult because this condition may be clinically indistinguishable from a variety of other intra-abdominal conditions such as appendicitis, inflammatory bowel disease, or other causes of small bowel obstruction [12]. This is particularly true in patients presenting with symptoms other than bleeding. In a study of 776 patients, 88% of patients presenting with bleeding had a correct preoperative diagnosis versus 11% of those with symptoms other than bleeding [16].

Plain radiographs are not usually helpful in making the diagnosis of MD. However, small bowel obstruction is usually visible on plain films of the abdomen. On CT, MD is difficult to distinguish from the normal small bowel in uncomplicated cases [2]. Although CT is being used more frequently to image the abdomen, the appearance of MD on conventional CT will vary according to the complication that precipitated the patient's presentation. In a report of CT findings in 11 patients with Meckel's diverticulitis, the presence of gangrene or secondary small bowel obstruction was associated with poorer diagnostic acuity. Administration of both intravenous and oral contrast material may help establish the diagnosis of Meckel's diverticulitis and should be administered whenever possible [17]. Finally, laparoscopy, as a diagnostic tool in cases of symptomatic MD, has also been reported [18].

Mortality in symptomatic patients is approximately 6% and higher in elderly patients with complications [11,18–20]. Delay in diagnosis of a complicated MD can lead to significant morbidity and mortality [9].

Surgical resection of symptomatic MD is the standard of care. Surgical options include simple diverticulectomy or ileal resection. The later procedure is preferred when there is evidence of severe inflammation, perforation or tumor [4]. Laparoscopic procedures can be performed without increased risk of complications by experienced surgeons [18]. Associated attachments to the abdominal wall should be removed. Cumulative incidence of early postoperative complications is 12%, including mainly surgical site infection (3%), prolonged ileus (3%) and anastomotic leak (2%) with a mortality rate of 1.5% [8]. Our patient underwent an ileal resection because of the presence of ischemic small bowel, and wound infection was the early postoperative complication observed.

4. Conclusion

In adults with symptomatic MD, the challenge presents itself in early diagnosis and prompt surgical treatment. Due to its rarity, high index suspicion is necessary as clinical presentation is variable, differential diagnosis is not straightforward and imaging techniques may not be useful. In young adults with small bowel obstruction, diagnosis is rarely made before surgery. To overcome these difficulties, CT scan with oral and intravenous contrast (if possible) is recommended. In our case, we retrospectively identified several risk factors that should have been identified to prevent delayed surgical intervention. Complications of a MD should be kept in mind in patients with atypical presentations.

Conflicts of interest

Nothing to state.

Funding

Nothing to state.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Table 1

| Topic | Item | Checklist item description | Reported on page |
|-------|------|-----------------------------|-----------------|
| Title | 1    | The words “case report” should be in the title along with the area of focus | Title Page |
| Keywords | 2    | 2–5 key words that identify areas covered in this case report | Keyword section |
| Abstract | 3a   | Introduction—what is unique about this case? What does it add to the medical literature? | 1 |
|   | 3b   | The main symptoms of the patient and the important clinical findings | 1 |
|   | 3c   | The main diagnoses, therapeutic interventions, and outcomes | 1 |
|   | 3d   | Conclusion—what are the main “take-away” lessons from this case? | 1 |
| Introduction | 4    | One or two paragraphs summarizing why this case is unique with references | 1 |
| Patient information | 5a   | De-identified demographic information and other patient specific information | 2 |
|   | 5b   | Main concerns and symptoms of the patient | 2 |
|   | 5c   | Medical, family, and psychosocial history including relevant genetic information (also see timeline) | 2 |
|   | 5d   | Relevant past interventions and their outcomes | 2 |
| Clinical findings | 6    | Describe the relevant physical examination (PE) and other significant clinical findings | 2 |
| Timeline | 7    | Important information from the patient’s history organized as a timeline | 2 |
| Diagnostic Assessment | 8a   | Diagnostic methods (such as PE, laboratory testing, imaging, surveys) | 2 |
|   | 8b   | Diagnostic challenges (such as access, financial, or cultural) | 2 |
|   | 8c   | Diagnostic reasoning including other diagnoses considered | 2 |
| Therapeutic Intervention | 8d   | Prognostic characteristics (such as staging in oncology) where applicable | NA |
|   | 9a   | Types of intervention (such as pharmacologic, surgical, preventive, self-care) | 2 |
|   | 9b   | Administration of intervention (such as dosage, strength, duration) | 2 |
|   | 9c   | Changes in intervention (with rationale) | 2 |
| Follow-up and Outcomes | 10a  | Clinician and patient-assessed outcomes (when appropriate) | NA |
|   | 10b  | Important follow-up diagnostic and other test results | 2 |
|   | 10c  | Intervention adherence and tolerability (How was this assessed?) | 2 |
|   | 10d  | Adverse and unanticipated events | 2 |
| Discussion | 11a  | Discussion of the strengths and limitations in your approach to this case | 3 |
|   | 11b  | Discussion of the relevant medical literature | 3 |
|   | 11c  | The rationale for conclusions (including assessment of possible causes) | 3 |
|   | 11d  | The primary “take-away” lessons of this case report | 4 |
| Patient perspective | 12   | When appropriate the patient should share their perspective on the treatments they received | NA |
| Informed consent | 13   | Did the patient give informed consent? Please provide if requested | Yes |
Author's contribution

Nâdia Tenreiro—study design, data collection, interpretation and writing.
Herculano Moreira—study concept, design and review of manuscript.
Sílvia Silva, Luis Madureira—data collection and interpretation.
João Gaspar, António Oliveira—review of manuscript.

Gurantor

Herculano Moreira.

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