Complicated Meckel's Diverticulum: lessons learnt from a case series of a single institution.

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Abstract

The aim of our study is to share a single pediatric institution's experience with complicated Meckel's diverticulum. The clinical records of a series of 15 cases treated between January 2006 and December 2020 were reviewed. Patients were divided according to the presenting symptoms (obstructive versus painful bleeding) and subcategorised according to demographic details, intraoperative findings, surgical technique, histological examination and length of stay. Patients older than 17 years of age were excluded from the study. Males were more prone to present with complicated Meckel's diverticulum than females, and obstructive symptoms were the most frequent presentation (60%). Median age at presentation was 9 years. The surgical technique of choice was an explorative laparoscopy in 12 cases, even though 25% of patients required conversion to laparotomy. There was no statistically significant difference between groups in terms of length of stay. Results are discussed with reference to current literature.

Introduction

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract. It is the result of an incomplete atrophy of the omphalomesenteric duct. The usual location of the diverticulum is on the antimesenteric border of the small intestine, most frequently between 30 cm and 90 cm from the ileocecal valve; there can be a fibrous connection to the umbilicus, a remnant of the partially obliterated vitelline duct [1].

MD is therefore a true diverticulum that affects approximately 2% of the population. It is often referred to with the 'rule of two': 2% of the population, within 2 feet of the ileocecal valve, 2 inches in length, two types of heterotopic mucosa, and presentation before the age of two.

Although it may be asymptomatic throughout their lifetime, MD carriers have a 2–4% risk of developing symptomatic complications [2].

The clinical manifestations in patients with symptomatic MD include acute abdomen caused by intussusception, ileus due to obstructive band, inflammation, perforation or severe anaemia (both acute and chronic) due to gastrointestinal bleeding [3, 4, 5].

The various clinical presentations make the diagnosis of MD challenging. In cases presenting with an acute abdomen, ultrasonography (US) or a plain X-ray is the preferred diagnostic method. In patients with acute or chronic intestinal bleeding from a suspected MD, abdominal US is the first line of examination, while esophagogastroduodenoscopy (EGDS) and colonoscopy under sedation are used to rule out high and low causes of bleeding [6]. However, as an acute bleeding from a MD is often severe and potentially life-threatening as well as an intestinal obstruction, diagnostic explorative laparoscopy has been advocated as the final diagnostic tool due to the inaccuracy of conventional imaging in diagnosing this condition [7].

This study aims to describe the clinical manifestation variety, the treatment of choice and the outcome in children with complicated MD referred to a single paediatric centre.

Materials And Methods

The clinical records of all consecutive cases younger than 18 years treated for complicated MD in a single paediatric centre (the Institute for Maternal and Child Health, IRCCS Burlo Garofolo) from January 2006 to December 2020 were retrospectively reviewed. Our definition of 'complicated MD' included all patients with clinical and/or radiological features of intestinal obstruction or peritonitis or severe anaemia (both acute and chronic) due to gastrointestinal bleeding, requiring surgical treatment. The final diagnosis was supported by intraoperative evidence of intussusception, intestinal obstruction, volvulus, perforation or gastrointestinal bleeding secondary to DM.

Medical records including clinical presentation at admission, preoperative diagnostics, intraoperative and histological findings, surgical approach, operative time, length of stay and postoperative complications were collected. Patients with a follow-up shorter than 6 months were excluded from the study.

The outcomes considered were length of hospitalisation and complications. Patients were then divided into two subgroups based on the symptoms at admission (abdominal pain versus rectal bleeding) and a comparison in terms of age, intraoperative findings, histology and outcomes was performed.

For categorical variables, Fisher's exact analysis was used. For continuous variables, Wilcoxon or Kruskal–Wallis tests were applied. Two-sided p values less than 0.05 were considered statistically significant.

Results

A total of 15 patients were included in this study (Table 1). This case series comprised 12 males (80%) and 3 females (20%) with a median age at presentation of 9 years (range 1–17 years). Intestinal obstruction was the most common complication, observed in 8 out of 16 patients (50%). The intraoperative findings of intestinal obstruction were intussusception in 4 cases (50%), presence of an obstructive band in 2 (25%) and volvulus in 2 (25%). All these patients presented with acute onset of abdominal pain and vomiting; the vomit was bilious in only one case. Six cases had a history of painless rectal bleeding with no other symptoms (40%). Gastrointestinal bleeding from MD was confirmed intraoperatively in all 6 cases, and in 2 cases the afferent artery to the MD was identified. Only one patient was admitted in our department for severe abdominal pain and fever, and diverticulitis was found intraoperatively.

All 15 patients underwent an abdominal ultrasound at admission. In 7/15 cases, paediatric radiologists were able to identify a tubular hyperechoic structure, raising the suspicion of MD, and in 85% of these patients (6/7) the ultrasonographic findings were associated with a bleeding MD, while the remaining case was associated with a kinking over a fibrous band starting from the tip of the MD. Regarding the other 8 cases, the 'bullseye sign' formed by a hypoechoic ring rounding an echo-dense centre (characteristic of intussusception) was found in 4 patients, while absence of peristalsis with paralytic ileus was found in 3 cases. Non-specific abdominal free fluid was detected in the only patient who showed a diverticular perforation with abscess. In 3 out of 4 cases with an ultrasound suspected intussusception, the level of the intussusception was unclear during sonography and an air contrast enema was attempted under sedation with midazolam. In all such cases, the pneumatic reduction was unsuccessful. In 2 patients with serious bleeding, EGD was performed in addition to abdominal US and no pathological findings were detected.

All patients underwent emergency surgery: 12 explorative laparoscopies of which 3 (25%) were converted to laparotomy for poor view and 3 were primary laparotomies. The median operative time was 90 minutes (range 60–250 minutes) without significant difference in the two groups ($p = 0.5$). In the laparoscopic technique group, there was no difference in the duration of the procedure even when performed by different surgeons (3 senior surgeons); however, the effects of a learning curve were observed, with a progressive decrease in the duration of surgery over the last 3 years. In 13 out of 15 cases (86.67%), a segmental small bowel resection including DM was performed, while in only 2 cases (15.38%) diverticulectomy alone was the procedure of choice.

Patients younger than 9 years were more likely to present with intussusception or volvulus than older patients, even though there was not a statistical difference ($p = 0.1$). The median MD length was 3 cm (range 1–5.5 cm) without statistical difference in the subgroups ($p = 0.78$). Histology confirmed the diagnosis in all cases. Ectopic tissue was revealed in 7 patients (46.6%): gastric tissue in 6 cases (85.7%) and pancreatic tissue in 1 case (14.3%). Comparing histological findings to symptoms and intraoperative evidence, ectopic tissue was more frequently found in patients with intestinal bleeding than in patients with abdominal pain ($p = 0.04$). Regarding postoperative complications, only one patient treated with the open technique developed an intestinal obstruction and underwent a second surgery after 35 days from the first intervention. The median length of stay was 5 days (range 3–16 days) without difference in the subgroups considered.

Discussion

MD is one of the most common congenital anomalies of the gastrointestinal tract, and was first characterised by Johann Friedrich Meckel. It becomes symptomatic in only 2–4% of cases, but its presentation can vary, making the correct diagnosis and the subsequent management difficult. For this reason, it is essential to identify patients at higher risk of complicated MD in order to diagnose and treat them promptly.

Generally, the current literature shows that the incidence of complicated MD is more common in males than females [8] with a variable rate from 2.3:1 [9] to 7.5:1 [10]. In the present study, the male-to-female ratio was 4:1, similarly to previous reported studies. A large proportion of complicated MD in childhood is reported to occur before 10 years of age [11] with the highest peak of occurrence under 5 years of age [4]. By contrast, in our case series, approximately half of the symptomatic cases (46.6%) were older than 10 years of age. We also found that the age distribution of patients in our cohort differed according to clinical presentation: patients with intestinal obstruction secondary to intussusception or volvulus were aged below 9 years in 5 out of 6 cases ($p = 0.1$), younger than patients with rectal bleeding who, in 67% (4/6) of cases, were 10 or older, although the difference was not statistically significant. This challenges the 'rule of two' stating that most complications, especially bleeding, occur before the age of 2. To the best of our knowledge, this is the first report that has examined the association between the clinical presentation and the age in children presenting with MD complications. Few other studies [8, 10] also reported a decreased frequency of complicated MD with age, but did not separately analyse each mode of clinical presentation and the age at onset of complications.

Traditional investigative modalities such as radiographic contrast studies, US, and computed tomography have many limitations in an accurate assessment of MD and its complications as they can mimic more common disorders that might cause an acute abdomen in children [12]. In case of bleeding episodes, a ^{99m}Tc -pertechnetate scintigraphy might be indicated; the principle is that a bleeding diverticulum consists of ulcerated ectopic gastric mucosa that can be revealed with ^{99m}Tc -pertechnetate. Indeed, it concentrates in the gastric tissue, leading to a reported sensitivity between 60% and 80% [13]. The problem is that, in cases of non-active bleeding, the sensitivity lowers further and, in any case, either a positive or negative examination would prompt a surgical exploration; for this reason, in our department, scintigraphy is not routinely used in cases of intestinal bleeding from a suspected MD.

Abdominal US has been considered the most effective tool to diagnose MD with high sensitivity and specificity [14]. MD is usually detected as a thick-walled tubular structure, with one blind end and the other end connected to the normal intestinal duct [15, 16]. Absence of peristalsis may allow the differentiation of MD from normal small bowel [17]. In our centre, abdominal US was performed routinely in all patients with acute abdominal pain and gastrointestinal bleeding. In a review by Higaki, it was reported that 88% of patients presenting with bleeding had a correct preoperative diagnosis, compared to just 11% of those with different symptoms [18]. This finding was confirmed in our study, in which more than 80% of patients presenting with gastrointestinal bleeding were properly diagnosed before surgery. However, in our series, abdominal US diagnosed up to 94% of patients correctly. This was likely due to the radiologist's expertise in a paediatric referral centre. We believe that healthcare professionals with multidisciplinary expertise might be useful to standardise the management and therefore to improve prompt diagnosis, especially in case of rare diseases.

For the treatment of symptomatic MD, surgical resection has always been the treatment of choice.

In our study, more than 80% of cases underwent a trans-umbilical laparoscopic-assisted (TULA) approach, in which the small bowel is exteriorised through the extension of the umbilical port site, as described by previous studies [12, 19].

The traditional procedure is diverticulectomy or segmental bowel resection and subsequent anastomosis, depending on the length of the MD, thickening or ischemia at the base, severe small bowel obstruction or intraoperative suspicion of incomplete resection of heterotopic mucosa [20]. Some retrospective studies have previously suggested that diverticulectomy can be considered an appropriate therapeutic strategy for bleeding MD [21, 22] or symptomatic MD without basal thickening or ischemia [23]. By contrast, in our series, diverticulectomy was only performed in 2 cases (1 bleeding DM and 1 abscess). Our centre's protocol includes a laparoscopic-assisted extracorporeal resection in case of complicated MD to ensure complete excision of any heterotopic mucosa and to verify intestinal vitality in case of intestinal obstruction. Postoperative complications after complicated MD resection are reported in literature to range from 5–12% in different series [24, 25]. In our study, the overall complications rate fell at the lower end of this range (5%) and the singular complication occurred after open surgery. Therefore, in our experience, TULA proved to be safe, feasible, and associated with low risks of postoperative complications even though it was not statistically significant, probably due to the small sample size. Postoperative histology revealed the presence of ectopic tissue from 41.1–85% in different series [26]. In our study, ectopic mucosa was confirmed in 46.6% of patients, with a higher

percentage in children with intestinal bleeding compared to those with abdominal pain ($p = 0.04$), in line with previous reports [24].

The last several years have seen some debate about the proper management of asymptomatic Meckel's diverticula incidentally found during laparotomy or laparoscopy. Tauro et al. suggested resection of a normal-appearing MD in every case of appendectomy or laparotomy/laparoscopy for an acute abdomen to avoid secondary complications [27]. Conversely, Robijn et al. suggested a selective approach, recommending resection of MD in certain cases: male sex, younger than 45 years, diverticulum longer than 2 cm, presence of a fibrous band highlighted at the time of surgery [28]. A recent systematic review by Zani et al. demonstrated that leaving an incidentally-found MD in situ reduced the risk of postoperative complications without increasing late complications [29]. As a result, our institutional policy avoids prophylactic DM resection as this practice is not supported by sufficient evidence.

Conclusions

The diagnosis of complicated MD in children remains a challenge. It should be considered in any patient with acute small-bowel obstruction with no history of abdominal surgery, especially if an intussusception or a volvulus is radiologically suspected. A complicated MD should be also considered in cases of GI bleeding. We advise bearing in mind that the age at presentation of patients with complicated MD is not always below 5 years, as the 'rule of two' would suggest. However, emergent exploratory laparoscopy is the procedure of choice in cases of unclear abdominal process. The trans-umbilical approach is a safe, effective and minimally invasive procedure to treat complicated MD.

Declarations

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with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from the individuals/legal guardians for the publication of any potentially identifiable images or data included in this article.

Authors' contributions: Conceptualization, J.S. and D.C.; methodology, M.D; software M.D and A.B; validation, M.D., D.C. and J.S.; formal analysis M.D; investigation M.D,M.M, D.O.; resources M.D, F.M and M.M.; data curation M.D and D.O.; writing—original draft preparation M.D and D.C; writing—review and editing D.C, E.G and A.B.; visualization M.G.S, E.G, G.DL, F.M; supervision J.S.; All authors have read and agreed to the published version of the manuscript. All the authors approved the final version of the manuscript and attest that they meet the ICMJE criteria for authorship.

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Table

Table 1. Summary of our cases.

Sex	Age (years)	Symptoms	US findings	Intraoperative findings	Surgical Approach	Surgical procedure	Histology	LOH days
M	14	Abdominal pain	Tubular hyperechoic structure	Inflammation + intestinal band	VLS à Open	Intestinal resection	Ectopic gastric tissue	7
M	7	Abdominal pain	Intussusception	Intussusception	TULAIR	Intestinal resection	Ileal mucosa	4
M	5	Abdominal pain	Paralytic ileus	Intestinal band	TULAIR	Intestinal resection	Ileal mucosa	5
F	10	Rectal bleeding	Tubular hyperechoic structure	MD bleeding	Open*	Intestinal resection	Ectopic gastric tissue	15
F	16	Rectal bleeding	Tubular hyperechoic structure	MD bleeding + afferent artery	TULAIR	Diverticulectomy	Ileal mucosa	5
F	15	Rectal bleeding	Tubular hyperechoic structure	MD bleeding + afferent artery	TULAIR	Intestinal resection	Ectopic gastric tissue	4
F	17	Abdominal pain	Free fluid	MD abscess	TULAIR	Diverticulectomy	Ectopic gastric tissue	4
M	1	Abdominal pain	Paralytic ileus	Volvulus	Open	Intestinal resection	Ileal mucosa	16
M	5	Abdominal pain	Intussusception	Intussusception	VLS à Open	Intestinal resection	Ileal mucosa	3
M	1	Rectal bleeding	Tubular hyperechoic structure	MD bleeding	TULAIR	Intestinal resection	Ectopic gastric tissue	3
F	13	Rectal bleeding	Tubular hyperechoic structure	MD bleeding	TULAIR	Intestinal resection	Ectopic gastric tissue	5
M	1	Rectal bleeding	Tubular hyperechoic structure	MD bleeding	TULAIR	Intestinal resection	Ileal mucosa	8
M	5	Abdominal pain	Intussusception	Intussusception	VLS à Open	Intestinal resection	Ectopic pancreatic tissue	7
M	13	Abdominal pain	Tubular hyperechoic structure	Volvulus	TULAIR	Intestinal resection	Ileal mucosa	6
M	9	Abdominal pain	Intussusception	Intussusception	Open	Intestinal resection	Ileal mucosa	5

TULAIR: Trans-Umbilical-Laparoscopic-Assisted-Intestinal-Resection

VLS: VideoLaparoscopic Surgery

*This patient developed an intestinal obstruction and underwent a second surgery 35 days after the first intervention.