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Case report

A case report of an asymptomatic necrotic Meckel's diverticulum in an inguinal hernia during elective surgery in a resource limited setting: Littre's hernia

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ABSTRACT

Introduction and importance: Although the common complications of Meckel's diverticulum (MD) are well known, that these congenital intestinal outpouchings may become involved as the content of abdominal hernia sacs is not well appreciated. MD is the most prevalent congenital abnormality of the gastrointestinal tract, but involvement in a hernia, known as Littre's hernia (LH), accounts for less than 1 % of MD cases. Incarcerated LH has been reported sporadically in the literature, with MD found in the sacs of paraumbilical, femoral, inguinal, and incisional hernias.

Presentation of case: We report a LH in a 3-year-old male child who was scheduled for elective herniotomy for a reducible left inguinal hernia. Intraoperatively we found the hernia sac contained a necrotic and perforated MD with viable associated bowel loop. The patient was successfully managed by diverticulectomy and primary repair through a trans-inguinal incision and herniotomy was performed.

Clinical discussion: LH is a rare presentation of MD, and preoperative diagnosis of LH is challenging. Even in the case of a strangulated MD, a patient may not present with the typical signs and symptoms associated with compromised viscous. Once identified, repair of Littre hernia consists of resection of the diverticulum, or segmental bowel resection if necessary, and herniotomy.

Conclusion: The finding of a perforated MD during elective hernia repair emphasizes the importance of awareness of unusual variants of inguinal hernia, and the necessity of identifying a MD given the risk of sequelae in the case of necrosis or perforation, if not repaired.

1. Introduction

Alexis Littre discussed the post-mortem findings of two cases in which “an appendix of the ileum “ had become incarcerated in inguinal herniae [1]. He defined this “new hernia,” and made the following relevant clinical observations concerning the pre-operative diagnosis: “(i) The patient goes to stool throughout the whole course of the illness, as the intestinal canal is uninterrupted. (ii) There is no vomiting, or by comparison, less frequently than with other herniae-it is never feculent. (iii) The belly is never fat, stretched or full with wind, as in ordinary herniae. (iv) The tumour (in the groin) is formed more slowly, and never becomes so large. (v) Inflammation, fever, pain or other symptoms which may accompany this peculiar kind of hernia, are less severe and take longer to manifest themselves than in other herniae [1].”

A Meckel's diverticulum (MD) of the gastrointestinal tract is a remnant of the embryologic vitelline duct occurring in approximately 2 % of the population [2]. It is typically found at the antimesenteric border of the ileum, usually located from 30 to 90 cm from the ileocecal valve containing gastric or pancreatic mucosa and remains asymptomatic in 91–96 % of people [3,4]. When symptomatic, individuals most commonly experience painless rectal bleeding, bowel obstruction, or inflammation [5,6]. The presence of an MD in any hernia space is termed a Littre's hernia. It has a reported incidence of 0.05 to 1 % of hernias, with approximately half presenting in inguinal hernias, and is rarely diagnosed in the pediatric population [7,8]. Its diagnosis is unlikely to be made before surgery [8,9]. Herein we report a rare case of asymptomatic, strangulated Littre's hernia during elective pediatric inguinal hernia repair at a private hospital in a low resource setting. This case is

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reported in line with the SCARE criteria [10].

2. Presentation of case

A 3-year-old male child presented to the pediatric surgical clinic in Uganda. His mother reported 10 months of inguinal swelling which was reducible upon lying supine. The child was consolable, not in distress or discomfort, with no signs of dehydration and normal vital signs. Physical examination revealed a 4 cm soft, nontender left inguinal mass that was reducible on lying. He had a soft non-distended non-tender abdomen with no guarding and no palpable mass or visceromegaly, and had no overlying skin changes. The mass was negative for transillumination. Hemoglobin and white blood count were 14 g/dl and 8.2 per mm^3 respectively and patient was scheduled for elective surgery.

Intraoperatively an edematous indirect inguinal hernia sac was noted, and this was dissected from the cord structures up to the level of preperitoneal fat. The hernia sac was then opened given the edema, and the content contained a localized necrotic and perforated tip of Meckel's diverticulum with viable associated bowel loop (Fig. 1). The content was mobilized out of the sac, the diverticulum measured 2 cm and was located on the antimesenteric surface and approximately 40 cm from the ileocecal junction. Interestingly, the hernia sac appears to have remained reducible and without free peritoneal contamination. This was because the neck of the sac was obstructed by grossly inflamed Meckel's diverticulum, and additionally, as on only the antimesenteric portion was involved, the symptoms remained subtle despite surprising intraoperative findings. Wedge resection of the diverticulum and primary repair was performed through the inguinal incision. Herniotomy was performed and the wound was closed in layers. Following identification of the contained perforated LH, intravenous metronidazole and ceftriaxone were given intraoperatively and the patient completed a total ten day course of antibiotics postoperatively. The patient made a rapid and uneventful postoperative recovery and was seen in follow-up at 6 weeks and remained well. The pathology report confirmed the presence of a large Meckel's diverticulum containing ectopic gastric and pancreatic mucosae.

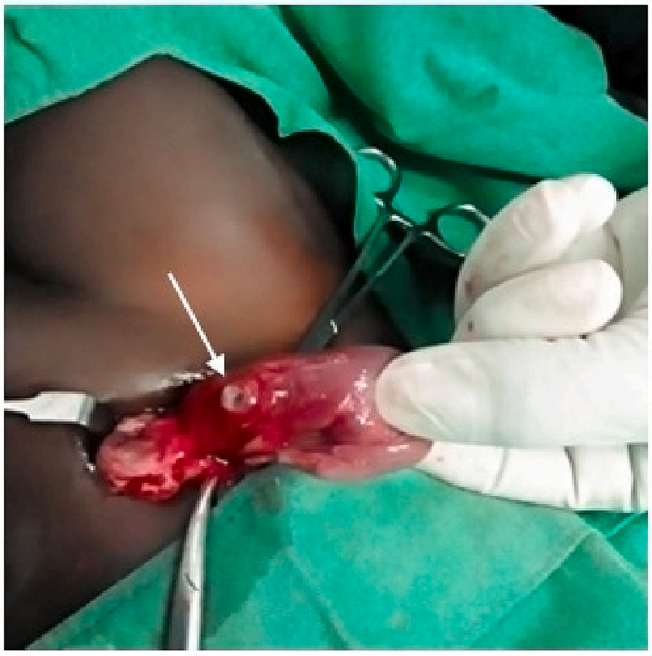

Fig. 1. Hernia sac containing Meckel's diverticulum. Perforated Meckel's diverticulum, with perforation at base of diverticulum indicated with white arrow, is noted within the opened hernia sac.

3. Discussion

MD is the most prevalent congenital abnormality of the gastrointestinal tract with an accepted incidence of 1 to 3 % [11]. LH is a rare presentation of a Meckel's diverticulum and represents less than 1 % of MD cases [8,11]. Among hernias, an estimated 0.05 to 1 % are found to be LH, most commonly discovered intraoperatively [8].

LH was first described by Alexis Littre in 1700 and at that time was attributed to traction [12]. Meckel's diverticulum, embryologically, is the persistent intestinal portion of omphalomesenteric duct or vitelline duct in which the midgut communicates with the umbilicus in utero till about five weeks of gestation. It is the most common congenital anomaly of the gastrointestinal tract [13,14]. It's usually found on the antimesenteric border of the ileum approximately 30 to 90 cm from the ileocecal junction measuring about 3 to 6 cm in length and 2 cm in diameter [3], as in the above case.

Fifty percent of the cases are found in the inguinal region, 12–30 % umbilical and 19–30 % femoral [3,8] It is more common in males than females i.e. male to female ratio is 3:1, and overall rare in children [15,16]. MD may be accompanied in the sac by the ileal loop to which it is attached. In children, it is mostly found in umbilical hernia, therefore making this case unusual, and the diverticulum is more prone to adhere to the sac [14].

Littre's hernia has been classified into two distinct subtypes. A "true" Littre's hernia, which is more common, contains the Meckel's diverticulum [13,16]. A "mixed" Littre's hernia contains a segment of small bowel in addition to Meckel's diverticulum and is less commonly reported [11]. This current case describes that of a mixed Littre's hernia, presenting with normal bowel but with asymptomatic necrotic Meckel's diverticulum [16,17]. Littre's Hernia is most commonly diagnosed incidentally during operative repair of inguinal hernia [16,17].

Despite the availability and wide use of modern imaging techniques, though not at easily obtained in resource limited settings, the diagnosis of LH remains elusive [14]. This case presents an infrequent complication, necrotic and perforated but asymptomatic MD in an inguinal Littre's hernia which only became evident during surgery [14].

Diagnosis of a strangulated Littre's Hernia was not made preoperatively as the presenting signs and symptoms were subtle and would typically evolve more slowly than those of strangulated small intestine, which has been previously reported [14]. A high index of suspicion is therefore needed to diagnose it correctly, and in this case the authors did not suspect the diagnosis preoperatively [18]. Fever, pain, signs of intestinal obstruction and peritonitis were not evident in this child, similar to a prior report by Muhammad S. Mirza and colleagues [14]. There may be no specific sign of bowel involvement other than local inflammation surrounding the hernia until an enterocutaneous fistula develops. In this case, edema of the hernia sac noted intraoperatively served as the only clue, and may have represented sequelae from a contained perforation. Obstruction can occur if the base of the diverticulum is broad enough to cause narrowing of the intestinal lumen which was not in this case [3].

Repair of Littre hernia consists of resection of the diverticulum and herniotomy [3], where the diverticulum is locally excised and bowel closed transversely, as was done in this case [13]. If the base of diverticulum is wide, as reported elsewhere, or the intestine appears nonviable, segmental resection of the involved loop of bowel with end-to-end anastomosis may be required [3,8,12,14]. This is then followed by repair of the femoral hernia [17,19,20]. Given the clinical presentation of narrow base and tip perforation of the MD in our case, resection of the diverticulum, primary repair and herniotomy was done.

Lastly, if one thought is to be left behind, it should be: "Meckel's is a great mimic that must be considered in all cases of intra-abdominal disease in which the cause is not readily apparent [16]".

4. Conclusion

We report an asymptomatic case of Littre's inguinal hernia with

necrotic and perforated MD. The symptoms and physical findings in this type of hernia are deceptively few and identification requires a high index of suspicion. Given the distal tip perforation of the MD in this case, diverticulectomy and primary repair through the inguinal incision as well as herniotomy was feasible. However, given the rarity of LH, the surgical approach should be dictated by the clinical presentation and the extent of the pathology encountered. This case emphasizes the importance of preparedness for unusual variants of inguinal hernia, and moreover, the necessity of identifying a MD given the risk of sequelae in the case of necrosis or perforation, if not repaired.

Ethical approval

No institutional approval was required for this case to be published rather this case was identified during routine clinical care.

Informed consent

Written informed consent was obtained from the patient's parents/legal guardian 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.

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Registration of research studies

Does not apply to this study.

Guarantor

Charles Odongo accepts full responsibility for the work and/or the conduct of the study, had access to the data, and controlled the decision to publish.

CRediT authorship contribution statement

All authors contributed equally in the literature search, interpretation of the article and review of the manuscript. All the authors have read and approved the final version of the manuscript.

Declaration of competing interest

The authors declare no conflict of interests.

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