



Littre's hernia: a case report of a rare intraoperative finding

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Background: Littre's hernia (LH) is a rare type of hernia defined by the containment of a Meckel's diverticulum. The true incidence of LH is unknown and has been mostly reported in the pediatric population, making the finding in adults infrequent and those located at the umbilicus exceedingly rare. Only 9 previous reports have described cases of umbilical LH. LH is largely asymptomatic making the diagnosis of these pre-operatively highly unlikely.

Case Description: We report the case of a 59-year-old man who presented for elective repair of an umbilical hernia. An intraoperative diagnosis of a LH was established. The lesion was subsequently repaired through a laparoscopic approach with partial bowel resection due to concerns for potential malignancy. To reduce the risk of infection, the use of mesh was omitted. However, based on low overall reported rates of surgical site infections in LH repair, mesh repair may represent a favorable alternative to primary closure alone.

Conclusions: Although exceedingly rare, LH should be included on the differential diagnosis of a newly discovered adherent mass during hernia repair surgery. Treatment options are diverse, and largely depend on operator preference. If the team elects to employ a mesh, recent literature suggests a pre-peritoneal approach is superior to an intra-peritoneal onlay.

Keywords: Case report; laparoscopy; Littre's hernia (LH); umbilical hernia

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Introduction

A Meckel's diverticulum (MD) is a remnant of the omphalomesenteric duct through which the fetal midgut communicates with the umbilical vesicle until the 5th week of gestation. The mucosa of these diverticula may contain gastric (23–50%), pancreatic (5–16%) or colonic tissue (1). Its reported incidence ranges from 0.6–4% making it the most common congenital anomaly of the intestinal tract (2). MD is most commonly clinically silent and present in 2% of the adult population. When symptoms are present, they can include gastrointestinal bleeding, bowel obstruction,

inflammation and/or perforation. Littre's hernia (LH) is a rare complication of MD in which a MD is contained within a hernia sac. Although MD is more common in men, LH is more common in women likely due to the higher incidence of obturator and femoral hernias. LH typically presents as an inguinal, femoral, or umbilical hernia. Previous reports have estimated a 1% incidence of LH among all cases of MD. While the true incidence of LH is unknown, only 67 symptomatic cases have been previously described in adults, with nine reports demonstrating LH localized at the umbilicus (3–23).

All but one of the case reports on umbilical LH are

pediatric, as the presentation in adults is unusual. It is generally poorly visualized by ultrasound or computed tomography (CT) and does not present with specific signs or symptoms (24). It is typically found intraoperatively during the hernia repair and confirmed by pathology. All symptomatic MD are typically resected but there is controversy regarding resection of incidentally found asymptomatic MD. Previously, reports describe single stage herniorrhaphy with or without diverticulectomy and/or small bowel resection as well as two-stage laparoscopic surgery as possible interventions for umbilical LH (4-11). We describe the case of an umbilical LH repaired laparoscopically, employing partial bowel resection including the MD and foregoing the use of mesh in a patient presenting for elective repair of a symptomatic umbilical hernia. We present the following case in accordance with the CARE reporting checklist (available at <https://ales.amegroups.com/article/view/10.21037/ales-22-45/rc>).

Case presentation

We present the case of a 59-year-old obese male [body mass index (BMI) of 40.87 kg/m²] with a previous medical history of hypertension, hypercholesterolemia, and type 2 diabetes mellitus who initially presented to the general surgery office with complaints of an umbilical hernia which had been increasing in size with worsening tenderness

for over 2 years. He reported no history of smoking or drinking, and there was no relevant family history. On physical examination, he exhibited tenderness overlying the umbilical hernia with reducibility. The patient denied obstructive symptoms such as changes in bowel movements, nausea, or vomiting. Additionally, he denied hematochezia on review of symptoms.

An elective laparoscopic umbilical hernia repair with mesh was scheduled. The patient was brought to the operative suite and placed in the supine position. Once adequate general endotracheal anesthesia was achieved, the patient was positioned appropriately with all pressure points protected. The abdomen was entered via a left upper quadrant incision at Palmer's point followed by insertion a transparent 12 mm trocar using a direct view technique in the mid upper abdomen. The abdomen was then insufflated to 15 mmHg pressure. Two additional ports were placed under direct vision: the first 10 mm port placed at the level of the umbilicus along the right anterior axillary line and a second 5 mm port placed on the left side of the abdomen at the same level.

A portion of ileum along with omentum was found herniating through the umbilical defect. Upon dissection of the hernia sac and moderate adhesions, a mass-like structure contiguous with the ileum was reduced from the hernia defect. Intraoperatively upon gross inspection, it was presumed to be a malignant mass versus an incarcerated MD, thus a 4–5 cm ileal bowel resection was undertaken. The specimen was removed through the umbilical defect after the skin and hernia sac were opened, partially excised and sent for pathology. The indication for small bowel resection was two-fold: one—due to clinical suspicion for a mass of unknown etiology and two—due to significant adhesions and concern for viability of this segment of bowel. The resected specimen can be seen in *Figure 1*. The entirety of the bowel was then inspected for other masses or lesions, but no other gross abnormalities were identified. Because of the need for bowel resection and escalation of wound class, we elected to proceed with a primary closure of the hernia and omit the use of mesh in attempts to mitigate infectious complications. The abdominal wall defect was approximated and closed under minimal tension with an absorbable monofilament suture. 0-vicryl suture was used to close the fascia of the 12-mm trocar site. The remaining port sites were irrigated and closed with 4-monocryl in a subcuticular fashion. An abdominal binder with a bandage with wicking action and absorbency, was placed over the hernia site to provide countertraction and reduce dead space. Pathology

Highlight box

Key findings

- A herniating mass continuous with the ileum may represent Littre's hernia, or a herniating Meckel's diverticulum. Symptomatic Meckel's diverticulum requires treatment with diverticulectomy, wedge resection, or ileal segment resection. Asymptomatic cases do not require surgical intervention.

What is known and what is new?

- It is known that Littre's hernia is the protrusion of a Meckel's diverticulum through an abdominal opening. This manuscript adds to the existing, but limited, evidence that Littre's hernia can complicate umbilical hernias. Further, this manuscript demonstrates the operative approach to resolving this type of hernia without the use of mesh. Lastly, this manuscript points out that Littre's hernia may be mistaken for a malignant mass.

What is the implication, and what should change now?

- Surgeons need to be aware of the diagnosis of Littre's hernia. Further, they must be prepared to tailor surgical treatment to each individual case, as treatments vary.

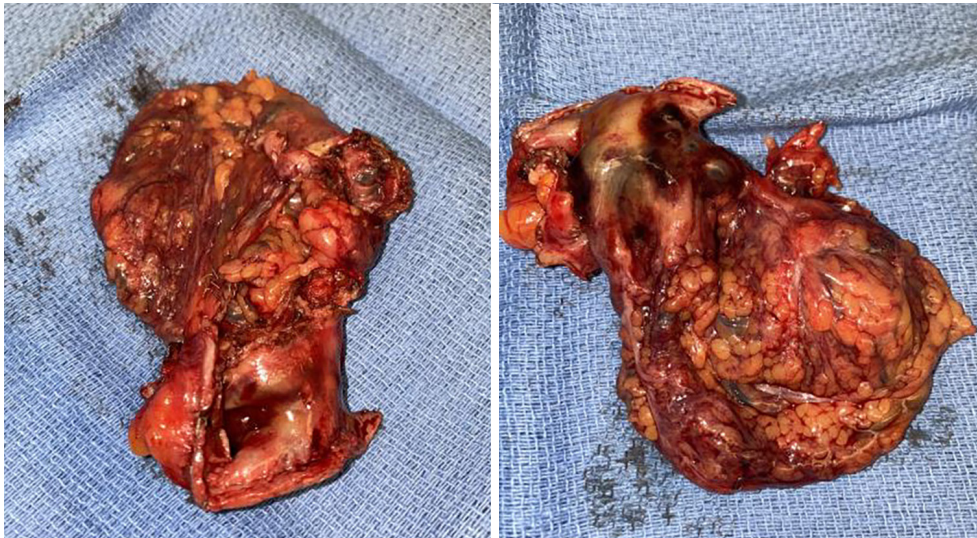


Figure 1 Gross resected ileum with abutting omentum and adherent mass like structure.

later confirmed that the resected bowel was indeed a MD, confirming an umbilical LH.

The patient recovered without postoperative complications and was discharged home on post-operative day one. The patient had no recurrence of symptoms during a 6-month follow-up period. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). 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 editorial office of this journal.

Discussion

MD is a congenital anomaly that is largely asymptomatic in 90% of cases. It is typically described by the “rule of 2s” in which the MD is seen in 2% of the population, diagnosed under 2 years of age, measures 2 inches in length, 2 cm in diameter, lies 2 feet proximal to the ileocecal junction, is 2× more common in males and is symptomatic in 2% of patients. Park *et al.* demonstrated in a review of 1,476 patients with MD—that when symptomatic, most children presented with obstruction while most adults presented with bleeding (25). Obstructive MD can occur by way of various mechanisms, including intussusception, volvulus around the umbilical attachment or incarceration/strangulation within a hernia. “The Mayo Clinic Experience” review identified four

risk factors predisposing to symptomatic MD in adults: age less than 50 years, male sex, diverticulum length greater than 2 cm and the presence of histologically abnormal tissue (25).

LH, however, occurs more frequently in women (60.4%) (26). LH occurs in the inguinal region 50% of the time, femoral 20%, umbilical and others 10%. Our case is rare as it describes an umbilical LH incidentally found during a routine umbilical hernia repair. Less than 10 previous umbilical LH have been reported in the literature.

Our patient was asymptomatic from the MD however, symptoms that can alert the surgeon to the possibility of a LH include a history of rectal bleeding, incomplete reduction or enterocutaneous fistula in a patient with a hernia (27,28). Most symptomatic cases present with non-specific symptoms of abdominal pain and occasional anorexia with malaise. Signs and symptoms of intestinal obstruction in LH are reported in 34% of cases (3). Technetium-99m scan can confirm the diagnosis pre-operatively but is often unnecessary and costly.

The management of MD is largely dictated by clinical presentation. If symptomatic, surgical resection is indicated. This can be done by means of diverticulectomy, wedge resection or ileal segment resection depending on the integrity of bowel and presence of ectopic tissue within the MD. When asymptomatic or incidentally diagnosed on imaging, foregoing surgical resection is preferred although optimal management remains controversial. In an epidemiologic, population-based study Cullen *et al.* recommended resection in patients younger than 80 years (29).

The paper published by the Mayo Clinic experience recommended removing all incidental diverticula that fulfill any of the 4 criteria identified to be associated with symptomatic MD (25). It remains unresolved whether diverticulectomy alone is sufficient in resection of MD versus a small bowel resection. Patients with ectopic tissue can have indirect involvement present as ulceration of small bowel adjacent to the diverticulum (30). There can also be involvement of the base of the diverticula. It is unclear whether retained ectopic tissue will become symptomatic (30).

Morbidity and mortality of surgical resection of MD is generally low as reported by the literature. In one small study the morbidity and mortality in a group of asymptomatic patients who underwent MD resection were found to be 20% and 3% (31). However, the complications did not correlate directly with the diverticulectomy itself. In our case presentation the Meckel's was resected as a safer alternative in the setting of a mass of unknown etiology and significant adherence to the surrounding bowel in this chronic umbilical hernia. Regarding the use of mesh, one systematic review of 53 cases of LH reported MD resection in all patients but the use of mesh in only 9 cases (17%), while the remaining underwent primary suture repair (3). Although it is unknown how many mesh repairs were complicated by mesh infection, only four patients experienced post-operative complications including wound dehiscence and minor surgical site infections (3). It is understood that the risk of field contamination was the primary factor for avoiding the use of mesh. It is important to note that most of the cases were reported prior to the 2000s. It is possible that data observing more recent LH repairs would reflect a higher use of mesh in patients without incarceration/strangulation or perforation.

The management of routine umbilical hernias largely relies on mesh to significantly reduce recurrence. Various techniques have been described for placement of mesh in ventral hernia repairs (32). Recent literature largely favors a pre-peritoneal approach to mesh placement versus an intra-peritoneal onlay approach due to increased cost-effectiveness, decreased post-operative complications and decreased recurrence (33,34). It is possible that this approach can be employed in cases of LH without significant adhesions or other technical factors prohibiting this. Due to the complexities of this case including the suspicion for malignancy, partial bowel resection and adhesions affecting the peritoneal planes, mesh was not utilized in the repair. Although the risk of hernia recurrence is higher without mesh, any potential complications of mesh placement were avoided.

Conclusions

In this case report, we identified a LH at the time of surgery for an elective umbilical hernia repair. Intra-operatively, the contents of the hernia sac included a small bowel mass of unknown etiology. It was resected along with a portion of bowel and subsequently returned as a MD on pathology. Less than 10 reports of umbilical LH exist within the literature. Presentation is non-specific and pre-operative diagnosis is thus difficult as in this case. The unknown etiology of the mass in this patient as well as the presence of adhesions and bowel resection prohibited the use of mesh in the umbilical hernia repair, although it is understood that this likely elevates the patient's risk for hernia recurrence. This unique case report presents the effective management of an incidentally discovered umbilical LH without the use of mesh. Follow-up will dictate further management as it pertains to hernia recurrence.

The patient was overall happy with the treatment they received. He reported gratitude for being able to get back to his normal routine without the discomfort associated with a symptomatic LH.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://ales.amegroups.com/article/view/10.21037/ales-22-45/rc>

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Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://ales.amegroups.com/article/view/10.21037/ales-22-45/coif>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised

in 2013). 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 editorial office of this journal.

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