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Case Report

Littré's hernia, a rare cause of intestinal obstruction: case report

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ABSTRACT

Littre's hernia is a rare condition in which a Meckel's diverticulum is found within a hernia sac, and it is an uncommon cause of intestinal obstruction. Preoperative diagnosis is challenging, and there is currently no consensus on optimal surgical management. Authors report the case of a 45-year-old Latin American woman who presented with intestinal obstruction due to an incarcerated umbilical hernia. Surgical exploration revealed a Littre's hernia, which was managed with stapled diverticulectomy without complications. The patient had an uneventful postoperative course.

Keywords: Littre's hernia, Meckel's diverticulum, Umbilical hernia, Incarcerated hernia, Intestinal obstruction, Case report

INTRODUCTION

Littre's hernia is a very rare type of hernia of which only about 100 cases have been reported in the literature.[1] Its preoperative diagnosis represents a clinical challenge, and the optimal surgical management remains controversial.[2,3]

Although there have been attempts to create algorithms for the proper treatment, currently there is a lack of consensus about the management in an incidentally found Littre's hernia, and in symptomatic ones there is still debate respecting the surgical technique (diverticulectomy vs segmental resection) and approach (open vs laparoscopic).[4-6] In accordance with the SCARE guidelines, we present the case of a middle-aged woman with intestinal obstruction, diagnosed intraoperatively with a Littré's hernia.[7]

The management, decision-making process and outcomes of this case are highly significant because of the scarcity of data regarding this rare pathology. The relevant current literature is going to be discussed and compared with the results of this case.

CASE REPORT

Patient information

A 45-year-old Latin American woman with a long-term history of umbilical hernia presented to the emergency room, with a 24-hour onset of symptoms, complaining of abdominal pain, umbilical bulging and vomiting of intestinal contents. A medical history of arterial hypertension treated with angiotensin-converting enzyme (ACE) inhibitors was reported.

As relevant past surgical interventions a meningocele repair in childhood and tumoral resection of a Schwannoma in the cerebellopontine angle with subsequent placement of a ventriculoperitoneal shunt thirteen years ago were also reported. As sequalae of the tumoral resection she presented with right side anacusis and facial palsy. Familiar and psychosocial history were deemed unremarkable.

Clinical findings

During physical examination it was found abdominal distention, augmented peristalsis, bulging at the umbilical region, with dimensions of 4 by 3 cm, without changes in skin color or local temperature, firm consistency and tenderness, content irreducible, painful at deep palpation predominantly in upper quadrants. After a nasogastric tube was placed for persistent emesis, approximately 1,500 ml of intestinal content were drained. Vital signs remained within normal parameters.

Diagnostic assessment

As a diagnostic approach in the emergency department, laboratory blood testing, plain abdominal X-ray, contrast-enhanced abdominal computed tomography (CT) scan and an abdominal Doppler ultrasound were performed. The laboratory findings show no significant abnormalities (Table 1). The plain abdominal X-ray showed small bowel distention up to 4.4 cm (Figure 1). The contrast-enhanced abdominal CT scan reported a 5.1 cm abdominal wall defect at the umbilicus, with protrusion of an ileum loop (Figure 2). The abdominal Doppler ultrasound reported an 18 mm wall defect at the umbilicus, a herniary sac of 56×23×39 mm, containing a bowel loop with adequate color Doppler signal (Figure 3). The physical examination, laboratory and imaging were consistent with the diagnosis of intestinal obstruction secondary to an incarcerated umbilical hernia. For this reason, the patient was admitted by the general surgery department for surgical management.

Table 1: Preoperative laboratory blood test results.

Laboratory blood test	Result	Reference range
Glucose (mg/dl)	129	70-105
Urea (mg/dl)	53.5	16.6-48.5
BUN (mg/dl)	25	6-20
Creatinine (mg/dl)	0.76	0.57-1.11
Sodium (mEq/l)	140.2	136-145
Potassium (mEq/l)	3.57	3.5-5.1
Chloride (mEq/l)	107.7	98-107
Calcium (mg/dl)	8.1	8.4-10.2
Phosphorus (mg/dl)	2.6	2.3-4.7
Magnesium (mg/dl)	1.9	1.6-2.6
Hemoglobin (g/dl)	13.1	13-18
Hematocrit (%)	39.5	42-53.6
Platelets (/µl)	238,000	150,000-450,000
White blood cells (/µl)	6,570	4,600-10,200
Neutrophils (%)	72.1	50-70
Prothrombin time (seconds)	11.3	9.4-12.5
International normalized ratio	1.04	0.8-1.1
Activated partial thromboplastin time (seconds)	32.4	25.1-36.5
Arterial blood gas		
pH	7.43	7.35-7.45
Partial pressure of carbon dioxide (mmHg)	29	35-45
Partial pressure of oxygen (mmHg)	87	80-100
Bicarbonate (mmol/l)	19.2	22-26
Lactate (mmol/l)	1.2	0.5-2
Base deficit (mmol/l)	-3.8	-4 to +2

Therapeutic intervention

Prior to intervention, consultation with the neurosurgery department was requested for assessment of the ventriculoperitoneal shunt. They determined that no further management was necessary if no contamination occurred. An urgent surgical intervention was performed the same day of presentation by an attending and residents of the General Surgery department. With signed informed consent, the patient was transferred to the operating room for laparotomy. Antibiotic prophylaxis with 1 g Ceftriaxone was administered 30 minutes before the procedure and a 12 cm supra-infra umbilical midline incision was made. Layer-by-layer dissection was performed until 6 by 6 cm hernia sac protruding through a 3 cm aponeurotic defect was found. The hernia content consisted of a 10 cm small bowel loop with macroscopically normal appearance and a 2×2 cm Meckel's diverticulum with erythema and edema (Figure 4). The aponeurotic defect was extended up to 4 cm to aid in the reduction of the small bowel loop back to the abdominal cavity. It was decided not to further explore

the cavity or manipulate the intestinal loops in order to reduce the risk of contamination and dysfunction of the ventriculoperitoneal shunt. Diverticulectomy was done using a linear stapler (Figure 5), with reinforcement of the staple line with running stitches using a 3-0 synthetic absorbable monofilament suture (Figure 6 and 7).

Aponeurosis repair was made with running stitches using a 1 synthetic non-absorbable monofilament suture, no mesh was placed. Subcutaneous tissue was approximated with simple stitches using a 3-0 synthetic absorbable multifilament suture and skin with simple stitches using a 3-0 synthetic nonabsorbable monofilament suture. The procedure ended without incidents.

Follow up and outcomes

The postoperative course went uneventful, managed as inpatient with intravenous NSAIDs and antibiotics with 1 gr Ceftriaxone b.i.d plus 500 mg Metronidazole t.i.d. only during hospital stay. Oral intake was initiated after retirement of nasogastric tube and assessment of passage of gas at postoperative day 3. The patient was discharged on postoperative day 7 with active mobility, adequate oral intake with normal diet and stool passage present.

The histopathology report revealed hyperplasia of mucosa-associated lymphoid tissue and vascular congestion. The patient was followed up after discharge with ambulatory visits on 1, 4 and 12 weeks. She exhibited appropriate healing of surgical wound, adequate oral intake, and no signs of pain, recurrence of intestinal obstruction, or signs of systemic inflammatory response.

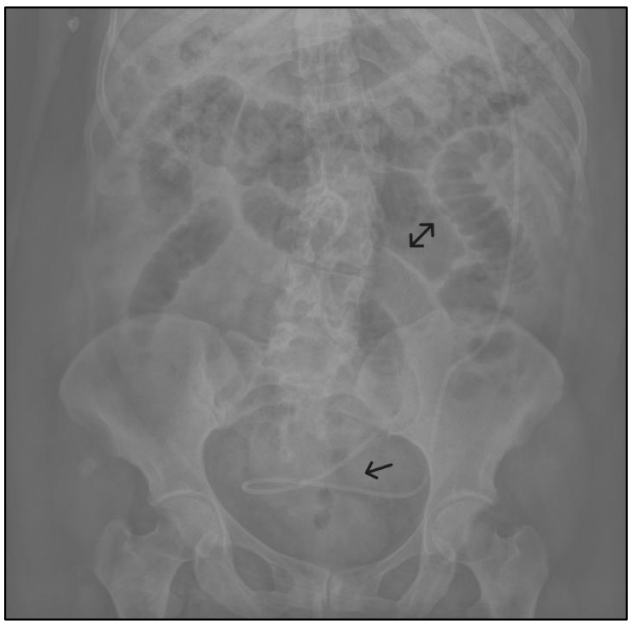

Figure 1: Abdominal X-ray. Preoperative abdominal X-ray showing small bowel distention (two-headed arrow); the ventriculoperitoneal shunt catheter can be seen (arrow).

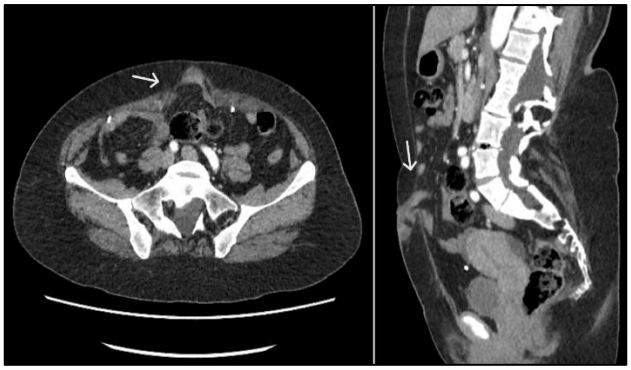

Figure 2. CT scan. On the left, an axial section of the preoperative CT scan showing an abdominal wall defect (arrow) at the umbilicus, with protrusion of an ileum loop. On the right, a sagittal reconstruction of the same CT scan.

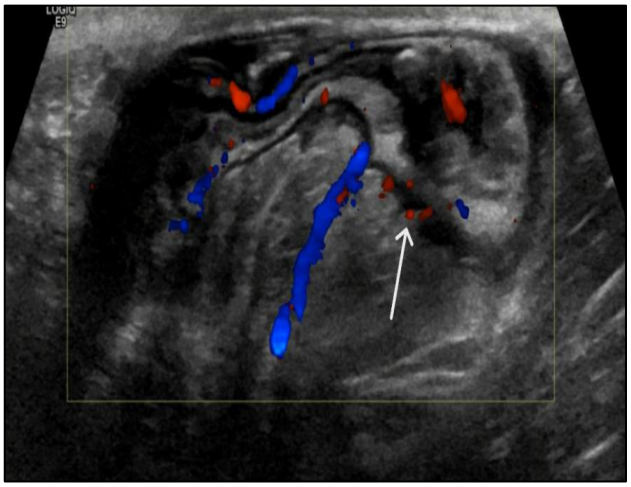

Figure 3: Doppler ultrasound. Preoperative abdominal ultrasound showing an umbilical defect with a small bowel loop as content (arrow), with adequate color Doppler signal.

Figure 4: Meckel's diverticulum. Meckel's diverticulum with its own arterial supply.

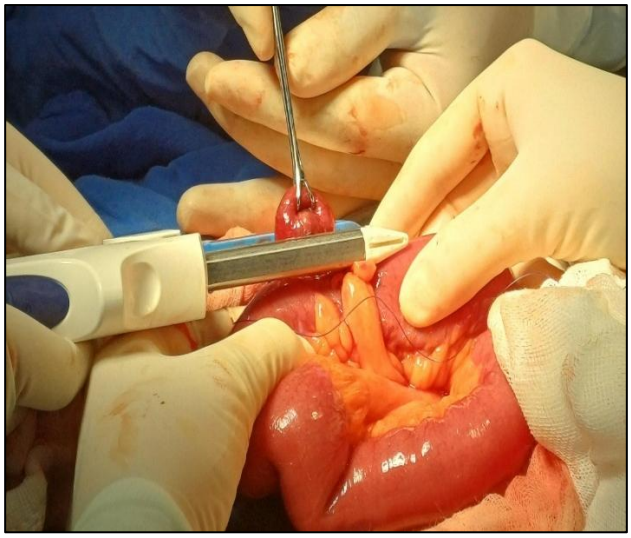

Figure 5: Stapled diverticulectomy. Diverticulectomy using a linear stapler.

Figure 6: Suture reinforcement. Ileum loop after stapled diverticulectomy and suture reinforcement.

Figure 7. Resected Meckel's diverticulum. Resected diverticulum next to a dissecting forceps.

DISCUSSION

The present report described a rare case of intestinal obstruction secondary to a Littré's hernia, of which only about 100 cases have been documented in the available English literature.[1] Littre's hernia was first reported in 1700 by the French anatomist Alexis Littre in his work titled Observation sur une nouvelle espèce de hernie. It was discovered in June 1699 during an autopsy and described as a conical-shaped appendix of the ileum, measuring four inches in length, within the sac of a left inguinal hernia.[8]

This "appendix of the ileum" had been mentioned earlier, in 1598 by Fabricius Hildanus and in 1671 by Lavater. However, it wasn't until a hundredth years after Littre's report, when the German anatomist Johan Friedrich Meckel recognized its embryological origin as a remnant of the omphalomesenteric duct in 1809.[9] The omphalomesenteric duct is an important structure in the early embryonic development that connects the yolk sac with the midgut. It is expected to obliterate and separate from the intestine between the 5th and 9th weeks of gestation. Anomalies in this process can result in vitelline duct pathologies, which include patent vitelline duct, umbilical sinus, vitelline cyst, fibrous cord and Meckel's diverticulum, the last being the most frequent.[10]

The Meckel's diverticulum is the most common congenital anomaly of the gastrointestinal tract, with a prevalence of 0.3 to 3% reported in the literature. It is a true diverticulum consisting of the three layers of the bowel (i.e. mucosa, muscularis and serosa) arising on the antimesenteric border and is typically found in the mid to distal segments of the ileum. It receives its own blood supply from the vitelline artery. Most of the patients are asymptomatic and are diagnosed incidentally during interventions for other reasons. Among symptomatic patients, most diagnoses occur during childhood, more often being male (with a male-to-female ratio ranging from 1.4:1 to 4:1) and typically under the age of.

The forms of presentation include gastrointestinal bleeding (the most common presenting symptom), obstruction, abdominal pain, nausea, vomiting or peritonitis due to perforation.[9-11] There's a classic mnemotechnic called the "Rule of twos" which states that the Meckel's diverticulum is present in about 2% of the population, 2:1 male to female ratio, located 2 feet proximal to the ileocecal valve, about 2 inches in length, 2-years old at presentation and two types of ectopic tissue in the mucosa lining (gastric and pancreatic).

According to a systematic review by Hansen et al the reported prevalence ranges between 0.3% and 2.9%, the male to female ratio from 1.5:1 to 4:1, the mean distance from the ileocecal valve was 52.4 cm, the length 0.4 to 11 cm and the diameter 0.3 to 7 cm, symptomatic presentation predominantly in children less than 5 years old. Most Meckel's contain ileal mucosa, but can also

contain ectopic tissue, 97% of all the ectopic tissue consisting of gastric or pancreatic tissue, the gastric tissue being present in 4.6% to 71% of symptomatic and 0 to 18.2% of silent Meckel's, and the pancreatic tissue in 0% to 12% of symptomatic and 0% to 5% of silent diverticula.[2] In a retrospective study by Tree et al including 160 patients, symptoms were associated with ectopic mucosa.[12]

The symptoms associated with Meckel's diverticulum are non-specific, making it hard to distinguish from other more common pathologies such as appendicitis. For such reasons, the preoperative diagnosis represents a challenge even for the experienced surgeon. Imaging studies including ultrasound, computed tomography, magnetic resonance and angiography can aid in the diagnosis, but their sensibility and specificity are low. However, they can support in the recognition of the cause of the manifestation (gastrointestinal bleeding, intestinal obstruction, intussusception, bowel perforation, volvulus). Endoscopic techniques such as double balloon enteroscopy and capsule endoscopy are modalities that can come to a preoperative diagnosis while approaching for lower gastrointestinal bleeding.[2,11]

A scintigraphy using Technetium-99 (Tc-99m) pertechnetate can be of great utility in preoperative diagnosis. It is often indicated in the context of obscure lower gastrointestinal bleeding in stable patients when other causes had been ruled out and the suspicion of Meckel's diverticulum persists. The scan localizes the diverticulum containing ectopic gastric mucosa trough uptake of the radiotracer. If a tagged red blood cell scan had been previously carried out, the Tc-99m scan should be performed 24 hours afterward, because the radiotracer may label the tagged red blood cells instead of the target ectopic mucosa. When properly performed, the Meckel scan has a sensitivity of 85% and specificity of 95%.[13]

The management of symptomatic patients requires surgical resection. On the other hand, the management of asymptomatic patients with intraoperative finding of Meckel's diverticulum remains controversial. It is estimated that about 4% of patients with Meckel's diverticulum would require hospital admission during their lifetime. Authors advocating against resection in incidental Meckel's diverticulum suggest that the risk of postoperative complications is higher in those who undergo resection, with no late complications in those left unresected.[3] A 2008 systematic review carried out by Zani et al reported a 5.3% postoperative complication rate in incidentally found Meckel's diverticulum patients who underwent resection vs 1.3% in those who were left in situ. In addition, they estimated a low mortality rate in asymptomatic patients, requiring 758 resections to prevent 1 death.[14]

A multicentered retrospective study by Tree et al from 1998 to 2017, including 160 patients who underwent resection of Meckel's diverticulum reported 92 symptomatic patients (57.5%) vs 68 asymptomatic patients (42.5%). The resection technique consisted of small bowel resection in 90 patients (56.3%), stapled diverticulectomy in 62 patients (38.8%), wedge resection in 7 patients (4.4%) and endo-loop diverticulectomy in 1 patient (0.6%). After a 12 month follow up, complications including bowel obstruction, chronic abdominal pain, pancreatitis, atrial fibrillation, reintervention for neuroendocrine tumor, incisional hernia and death (due to pulmonary embolus and hemorrhagic pancreatitis) were encountered in 15 patients (9.4%), 11 (73.3%) in the small bowel resection group and 4 (26.6%) in the diverticulectomy group. This demonstrated a higher complication rate in patients who underwent small bowel resection (p=0.044), however, a subgroup analysis by demographic characteristics was not carried out.[12]

There have been intents of creating algorithms to aid in the decision-making process for Meckel's diverticulum. Blouhos et al proposed that the type of resection depended on the integrity of the diverticulum base and adjacent ileum and the presence and location of ectopic tissue. The location of the ectopic tissue can be predicted based on the height-to-diameter ratio (long diverticula height-to-diameter ratio >2). In long diverticula, the ectopic tissue distribution is at the body and tip, while in short diverticula, the ectopic tissue distribution includes the base. They proposed that for short diverticula, wedge or segmental resection should always be performed, whereas for long diverticula, simple diverticulectomy can be performed except in cases of complicated diverticulitis or complicated intestinal obstruction, in which cases the wedge or segmental resection are preferred.[4]

The outcomes of laparoscopic vs open resection are comparable in terms of operative time, length of stay and postoperative complications. A 2018 retrospective comparative study in pediatric population carried out by Ezekian et al comparing laparoscopic (N=73) vs open approach (N=75) reported an operating time of 71 vs. 65 minutes (p=0.187) and a total length of stay of 4 vs 4 days (p=0.178) respectively. Complications including surgical site infection, organ/space infection, pneumonia, bleeding, reintubation, reoperation and readmission were comparable between the two groups, with a p value >0.05 in all terms. A rate of conversion from laparoscopy to open approach of 27.4% was also reported.[5]

Another 2020 retrospective study by Skertich et al compared laparoscopic (N=267) vs open approach (N=295) in pediatric patients. The operative time was 71.59 and 76.57 minutes respectively (p=0.449) and the total length of stay was 3 and 4 days respectively (p=0.009). Morbidities including bleeding, pneumonia, unplanned intubation, surgical site infection, wound dehiscence, organ/space infection, sepsis and urinary tract infection were similar between groups, 10.5% vs 16.6% (p=0.164).[6] As stated before, a Littré's hernia is a Meckel's diverticulum inside a hernia sac. It can occur at

an inguinal, femoral, umbilical, obturator, Spigelian or ventral defect, being the inguinal the most common site. It is a rare finding during surgery, which exact incidence remains unknown, but it is estimated that about 1% of patients with a Meckel's diverticulum will subsequently present with a Littré's hernia.[15] Since the symptoms are the same as those of a regular hernia, it is usually diagnosed intraoperatively. It is important to distinguish it from a Richter's hernia, where only a part of the intestinal wall is incarcerated at the herniary orifice.

A true Littré's hernia is that which contains only the diverticulum, whereas a mixed Littré's hernia is that containing the diverticulum and another portion of small intestine or other organ. It is estimated that the incidence of a true Littré's hernia is 7 times that of a mixed one. Intestinal obstruction may not be present in a true Littré's hernia, because only the diverticulum is inside the hernia sac and the bowel loop remains free.[16] Although Meckel's diverticulum is more common in men, Littré's hernia is more common in women, representing about 60.4% of cases, most likely because of the high incidence of femoral and obturator hernias.[15] However, systematic review by Racareanu et al stated that the male-to-female ratio from available data was almost equal.[1] The standard treatment consists of the hernia repair and management of Meckel's diverticulum through diverticulectomy or segmental resection if compromised or left in situ if it was incidentally discovered.[16]

CONCLUSION

The case presented in this document is relevant because of the low incidence of Littre's hernia reported in the literature, the controversy regarding the management when it is incidentally found and the lack of consensus with respect to the surgical technique, diverticulectomy vs resection. In this particular scenario, although the height-to-diameter ratio was <2, it was decided to perform simple diverticulectomy in an attempt to reduce the risk of contamination of the abdominal cavity because of the patient antecedent of ventriculoperitoneal shunt.

The pathology report revealed that no ectopic tissue was found in the diverticulum, in accordance with the previously discussed fact that ectopic tissue is mostly found in a symptomatic Meckel's diverticulum as compared to silent diverticula. The postoperative course went uneventful, supporting the low incidence of diverticulectomy-associated complications reported in other studies cited in this paper. This case highlights the decision-making process in complex cases of Littre's hernia, such as those involving a ventriculoperitoneal shunt and emphasizes the need for individualized treatment approaches. Case series and reports like this one on Littre's hernia are important because further research is needed to establish evidence-based recommendations and consensus regarding diagnosis, management and optimal surgical technique.

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