

Littre's hernia: a case presentation

Hernia de Littre : presentación de un caso

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Abstract:

It is reported a case of Littre's hernia in a 64-year old woman, which is a very uncommon condition, consisting on an hernia whose content corresponds to a remnant of the omphalomesenteric duct, called Meckel's diverticulum, that results from incomplete obliteration of the vitelline duct between the fifth and sixth weeks of human gestation as the bowel settles into its permanent position within the abdominal cavity. The diagnosis was made from the clinical findings and supported by diagnostic auxiliaries such as ultrasound and computed axial tomography (CAT). The intervention performed was a laparoscopic transabdominal pre-peritoneal inguinal repair (TAPP). The fact that this condition was diagnosed until the sixth decade of age and that the patient was a woman enhances the rarity of this case.

Keywords:

Littre's hernia, Meckel's diverticulum, incarcerated hernia.

Resumen:

Reportamos un caso de hernia de Littre en una mujer de 64 años de edad, lo cual es una condición muy poco común, consiste en una hernia cuyo contenido corresponde al remanente del conducto onfalomesentérico, llamado divertículo de Meckel, resultado de una incompleta obliteración del conducto vitelino entre la quinta y sexta semana de gestación mientras el intestino se asienta en su posición permanente en la cavidad abdominal. El diagnóstico fue realizado a partir de los hallazgos clínicos apoyado de auxiliares diagnósticos como ultrasonido y tomografía axial computarizada (TAC). La intervención realizada fue una reparación inguinal laparoscópica transabdominal pre-peritoneal (TAPP). El hecho de que esta condición fuera diagnosticada hasta la sexta década de la vida y que la paciente perteneciera al sexo femenino hace más inusitado el caso.

Palabras Clave:

Hernia de Littre, divertículo de Meckel, hernia encarcelada.

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INTRODUCTION

A Littre's hernia is a hernia that contains an incarcerated Meckel diverticulum. A diverticulum is a blind pouch that is extruded from the gastrointestinal tissue. It can occur at any level within the gastrointestinal tract, from the esophagus to the colon (where it is more common 50%). Diverticula are classified according to their histologic and etiologic characteristics in two types: Meckel and non-Meckel diverticulum. Histopathologically, Meckel's diverticulum includes the invagination of the mucosa, submucosa, muscular and external serosa; on the other hand, the non-Meckel's diverticulum only includes the invagination of the mucosa, submucosa and serosa through the muscle at the vessel-penetration sites.^{1,2}

It has been estimated that the prevalence for the Meckel's diverticulum is about 2% of the general population.^{1,2} It is the most common congenital anomaly of the gastrointestinal tract, and it is derived from the persistence of the omphalomesenteric tube, due to an incomplete obliteration of the vitelin duct during the fifth week of gestation.² In general, it persists without clinical manifestations in the form of a diverticulosis, suddenly inflaming to produce a diverticulitis, which has no pathognomonic symptoms or signs.³

With regard to the anatomy and embryology of Littre's hernia which contains Meckel's diverticulum, the anatomic formation of the intestine is achieved since the fourth fetal week. Each step of development can be influenced by a variety of processes, including microbiome, genetic, cellular, neural, and hormonal regulatory mechanisms.⁴

Although Meckel's diverticulum usually remains clinically silent, a logistic regression analysis of 1476 medical records with Meckel diverticulum made by Mayo Clinic from 1950 to 2002 shows that only 16% were symptomatic and that the most common clinical presentation in adults was bleeding whereas in children, obstruction. Not only but also, they found increased risk of developing symptoms in patients younger than 50 years old, male sex, with a diverticulum length greater than 2 cm and presence of histologically abnormal tissue.⁵

Although there are multiple diagnostic methods, the gold-standard diagnosis method for the Meckel diverticulum when gastric ectopic tissue is suspected clinically by digestive tract bleeding is called Meckel's scan and it consists in gammagraphy with tecnecio 99 just because this radiopharm has a lot of affinity for the gastric mucosa, its sensivity had been calculated to 85-97%.⁶

Moreover, arteriography, wireless capsule endoscopy and double-balloon enteroscopy can be used too.² As we know hernia's diagnosis is based on clinical examination, and mostly, it is made intraoperatively during abdominal exploration performed for another prescription or even on exploratory laparotomy.^{1,7}

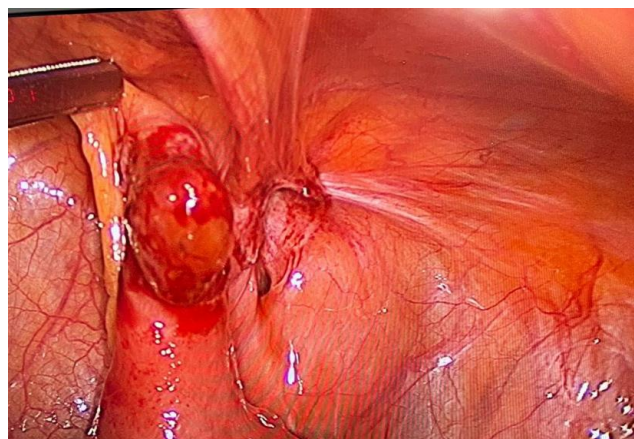


Figure 1. Transoperative finding of the Littre's hernia. The content of the hernia was visualized during the surgery.

Diverticulitis-derived complications can range from gastrointestinal bleeding and obstruction to perforation.⁸ Although many authors agree that the most common complication in both adults and children is gastrointestinal hemorrhage, a logistic regression analysis made by Mayo Clinic, consisting on 1476 medical records from 1950 to 2002, showed that obstruction was the most common clinical presentation.⁸ Gastrointestinal bleeding can present as painless rectal bleeding children, without further symptoms or signs of gastroenteritis, and in adults diverticulitis is suspected when gastrointestinal bleeding with an unknown source is identified.^{9,10} Furthermore, a systematic review of forty-five studies reported 53 patients (21 males and 32 females) with Littre's hernia, found that most common sites of occurrence were femoral (21 patients, 39.6%) and inguinal (18 patients 34%), whereas 77.4% concerned incarcerated hernia.¹⁰

An increased risk of developing symptoms occurs in patients younger than 50 years old, of male sex, and with a diverticulum length greater than 2 cm with the presence of histologically abnormal tissue.⁸ Interestingly, 43% of symptomatic patients present with ectopic tissue, and the most common ectopic tissue found in these cases has a gastric origin (33%).⁸ In this case, the ectopic gastric mucosa within the diverticulum produces acid secretion that causes adjacent ulceration downstream the diverticulum.

Although uncommon, the prevalence of the Meckel's diverticulum has already been studied. For instance, a nationwide population-based study in Taiwan shows that the annual incidence was 28.55 per 100,000 person-year rate between 1996 and 2013, in which only 260.5 per 100,000 person-year were symptomatic.¹⁰ Moreover, in a retrospective study recruiting 825 patients' files, 1.45% was detected to suffer from Meckel's diverticula by analyzing the videos of capsule endoscopy studies.¹¹ Finally, a systematic review addressing eight studies that in sum report the condition of 56,722 patients (all undergoing appendectomy, with Crohn's disease or bodies

in which necropsy was performed) was used by us to average the prevalence of the Meckel's diverticulum, which was calculated to be $1.54\% \pm 0.87$, which suggest that the pathology is sparse within the worldwide population.¹² Despite this, the Littre's hernia is so uncommon and tends so much towards being silent that a method to establish the prevalence of such an interesting entity hasn't been developed, but it must have an enhanced rarity over that of the Meckel's diverticulum.

As we know the treatment for hernias is surgical repair, but a hernia case turns critical and needs urgent surgical intervention in the context of acute incarceration or strangulation.¹³ In this case, the treatment option that was chosen involved the minimally invasive repair, being the transabdominal pre-peritoneal patch (TAPP) repair unassisted hand, whose recurrence calculated by a meta-analysis which includes 5 studies made by the Instituto Mexicano de Neurociencias, Facultad de Ciencias de la Salud, Universidad Anáhuac México found that there was 14 cases of recurrence between 275 patients which rises up to 5% of recurrence.^{14, 15}

In contrast, a retrospective study made by the Hospital Militar de Mexico between April 2017 and December 2019 did not found recurrence.¹⁶

Moreover, in addition to the imperative indication of surgery due to the incarcerated inguinal hernia, for female patients with a groin hernia, surgical intervention is recommended regardless of symptoms or the type of hernia. The reason is that females are four times more likely to have femoral hernias than males, and femoral hernias are eight times more likely to strangulate than inguinal hernias (36 to 39 versus 5 percent).^{12, 17, 18}

In contrast, watchful waiting could be performed in males with asymptomatic inguinal hernias and pregnant patients with uncomplicated inguinal hernias. Nonetheless, it is necessary to avoid modifiable risk factors, including smoking cessation and weight loss.¹⁹

CASE REPORT

This case report was made according to Helsinki Declaration and was approved by the Ethics Committee of the Sociedad Española de Beneficencia. The patient in question authorized the use of her data for the elaboration of this article through informed consent.

A 64-year old woman was admitted to the Hospital Español de Pachuca, in Hidalgo, Mexico with a painful inguinal swelling after fifteen days of evolution. Upon physical examination a flat, depressible and painless abdomen with adequate peristalsis was detected; nevertheless the presence of non-reducible and painless to mid and deep palpation bulge of about 2x2 cm in the right groin area was remarkable. This finding led to the diagnosis of incarcerated inguinal hernia. She already had performed an USG that reported an elliptical anechoic image with a hypoechogenic

ring of approximately 5 x 5 mm, without evidence of inflammatory adenopathies. Overall the information was suggestive of an incarcerated inguinal hernia that does not get bigger with maneuvers that increase intra-abdominal pressure. Furthermore, an abdominal TAC was also performed where a right inguinal hernia of intestinal content was appreciable. Due to the high risk of ischemia and intestinal necrosis, or need for intestinal resection and subsequent anastomosis, urgent surgery was decided.

As a surgical history, the patient had an open resection of ovarian cyst 30 years ago, hysterectomy secondary to myomatosis in elliptical image 13 years ago and two ocular surgeries 24 and 18 years ago.

The surgical plan consisted in a laparoscopic transabdominal pre-peritoneal (TAPP) inguinal hernia repair not hand-assisted. At surgery, a Meckel's diverticulum was found to be the inguinal hernia's content, which is likely to have distal vascular compromise, the hernial sac was localized, reduced, and the peritoneum was incised in order to perform the peritoneal flap. The rest of the ileus was explored in order to ensure lackness of another anatomic alteration or vascular compromise. Then, at the visualization of Cooper's ligament, it was dissected through the Retzius space and the Bogros space was dissected as well. The vision of the critical area of the myopectineal orifice was achieved, polypropylene mesh was placed and sutured with polypropylene 2-0, and the peritoneal flap was closed with polypropylene 3-0 before doing enterotomy with latero-lateral anastomosis of 60mm in order to resect the Meckel's diverticulum. Enterotomy was closed with ethinbond 2-0 (Figure 1).

To ascertain the absence of intestinal leakage it was used methylene blue without getting blue expense through Penrose, only serous output.

The pathology department reported a Meckel's macrodiverticulum of 4 x 3 x 2 cm weighing 11 grams, with an unpolished serosa with congestive vessels, and hemorrhagic and adenomatous mucosa with thickened folds.

Histological cuts indicated the presence of shortened intestinal villi, enhanced cellularity of the lamina propria, light edema and moderate inflammatory infiltration composed of neutrophils, lymphocytes and plasmatic cells. Tissue's vascularization was shown to be congestive with focal extravasation of erythrocytes and necrotic plaques (Figure 2).

DISCUSSION

Although hernia's repair is one of the most commonly performed operations reaching the total of 20 million inguinal or femoral hernias repaired every year worldwide, only a 2% prevalence is estimated for the Meckel's diverticulum in the community, and thus it was thought that the Litter's hernia prevalence must be much lower, but impossible to determine with current data.^{4, 20}

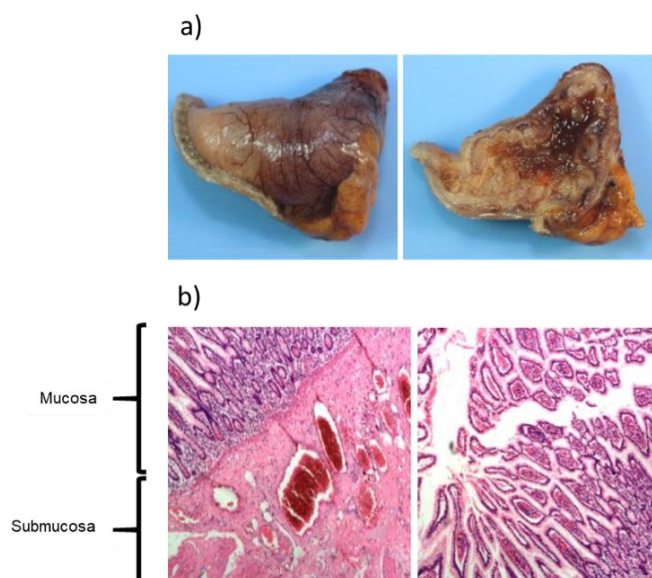


Figure 2. Resected Littre's hernia. Macroscopic examination of the resected tissue (a) as well as microscopic

Interestingly, the “rule of twos” is commonly used to describe the frequency of the Meckel's diverticulum characteristics, corresponding to 2% of the population being affected with a 2:1 male-to-female ratio, and the location of the diverticulum which is usually two feet from the ileocecal valve as well as a common length of two inches.²¹ Thus, in addition to the uncommon nature of a Littre's hernia by itself, another rarity in this case is the fact that the treated patient was a woman. Moreover, Meckel's diverticulum is usually clinically silent, and its Littre's herniation must be its most uncommon presentation.^{22,23}

Particularly in this case it was appreciated a rare presentation because of the findings in the physical examination, due to unreducible painless mass to mid and deep palpation, there was no characteristic symptom or sign that could guide to the diagnosis of a Littre's hernia. Moreover, imaging studies like ultrasonography and tomography didn't show any additional information that could guide the diagnosis. However, clinical findings lead to the diagnosis of incarcerated hernia, but its confirmation was intraoperative.

CONCLUSIONS

Despite the uncommon condition of Littre's hernia, clinical awareness is important in the context of incarcerated hernia or gastrointestinal bleeding without identifiable source, however it should be considered as a differential diagnosis.

In this case it was showed that the clinical presentation of Littre's hernia is versatile and nearly always is identified incidentally or during surgery; however, once identified and as it is requested, depending on the clinical conditions and the individual characteristics of each person, the ileal resection of Meckel's

diverticulum allows the removal of all ectopic tissue and avoids complications.

CONFLICTS OF INTEREST

None of the authors have any conflict of interest to declare.

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