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A rare case of a strangulated Littre's hernia with Meckel's diverticulum duplication. Case report and literature review

C.R. López-Lizárraga^{a,*}, M.P. Sánchez-Muñoz^b, G.E. Juárez-López^c, L. Pelayo-Orozco^d, L.F. De la Cerda-Trujillo^e, C.F. Ploneda-Valencia^{e,*}^a Division of Surgery, Hospital Civil de Guadalajara "Dr. Juan I. Menchaca", Salvador Quevedo y Zubieta street #750, Guadalajara, Jalisco, Mexico^b Department of Bariatric Surgery, Hospital Civil de Guadalajara "Dr. Juan I. Menchaca", Mexico^c Department of Pathology, Hospital San Javier, Guadalajara, Jalisco, Mexico^d Department of Surgery, Hospital IMSS, 180 Guadalajara, Jalisco, Mexico^e Department of General Surgery, Hospital Civil de Guadalajara "Dr. Juan I. Menchaca", Mexico

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ABSTRACT

INTRODUCTION: The Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract present in approximately 1–4% of the population; the MD duplication is exceedingly rare with only a few reports of it. Here we present the first case of a strangulated Littre's hernia with MD duplication.

PRESENTATION OF CASE: A 30-year-old male presented to the emergency room with clinical signs of small bowel obstruction, at physical examination, a right incarcerated inguinal hernia with erythema was found. We did a laparotomy, and two MD were found, one in the sac with ischemia, and the other 90 cm from the Bauhinis valve. A diverticulectomy of the ischemic diverticulum was done, and the other MD was left in place; the inguinal region was repaired with a Lichtenstein technique.

DISCUSSION: The complications of the MD are 3–4 times more frequent in men, been an intestinal obstruction, hemorrhage, diverticulitis, ulceration, and perforation. A Littre's hernia is when the MD is found in the sac; this is seen in the inguinal region in 50% of the cases. The management of a Littre's hernia is the resection of the MD; it could be done by an intestinal resection or by a diverticulectomy accordingly to the Park criteria.

CONCLUSION: As to our knowledge, this is the first case of an incarcerated Littre's hernia with duplication of a Meckel's diverticulum.

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1. Introduction

Meckel's diverticulum (MD) is the most common congenital anomaly of the gastrointestinal tract, occurring in approximately 1–4% of the population [1–4]. Johann Friedrich Meckel described the anatomy and embryology of the diverticulum in 1809, and Alexis Littre identified the presence of the diverticulum in the sac of a hernia in 1700 [1]. The MD is usually asymptomatic, been complicated in 5–17% [4,5]; the most frequent complications in adults are an obstruction (14–53%), diverticulitis (12.7–53%), ulceration (<4%), and perforation [3,6]. Duplication of the MD is exceedingly rare, with only a few reports of it [4–12]. Here we present the case of a strangulated Littre's hernia with a duplication of a Meckel's diverticulum, as to our knowledge, this is the first case with this unusual presentation. The following accomplishes the SCARE criteria [13].

* Corresponding authors.

E-mail addresses: lopezlizarraga43@yahoo.com (C.R. López-Lizárraga), plonecef@gmail.com (C.F. Ploneda-Valencia).

2. Case presentation

A 30-year-old man presented to the emergency room with nausea, feculent vomiting, and a painful mass with erythema in the right inguinal region. At physical examination, the patient had a distended abdomen with diminished peristalsis. The abdomen was tenderness with Blumberg present. The right inguinal region had an erythematous soreness mass not reducible. The abdominal x-ray showed a small bowel obstruction (Fig. 1). A laparotomy was performed.

A midline incision was done, we found the distal ileum inside the inguinal ring, and once the adhesions were released, a strangulated Meckel's diverticulum was seen at 20 cm from the Bauhinis valve, it was 3–4 cm in length without any palpable mass in it (Fig. 2). A Noble maneuver was done, and another MD was seen at 90 cm from the Bauhinis valve with a length of 1.5–2 cm without a palpable mass in it (Fig. 2). The ischemic ileum regained its normal coloration and blood flow (Fig. 3); hence, a diverticulectomy was performed. The inguinal region was repaired with a Lichtenstein technique.



Fig. 1. Abdominal x-ray showing dilatation of the small bowel.



Fig. 2. Meckel's diverticulum duplication.



Fig. 3. Meckel's diverticulum with ischemia.

The patient had no postoperative complication and was discharged on the fourth postoperative day. The histopathology reported a Meckel's diverticulum with ischemic enteritis, and ectopic gastric mucosa. After six months from the surgery, the patient has no complained or abdominal discomfort.

3. Discussion

The MD was first described by Fabricius Hildanus in 1598, and later by Lavater in 1691, but was Johann Friedrich Meckel who described the anatomy and embryology of the diverticulum in 1809; that is the reason for the diverticulum name [1,3,9,10]. The Meckel's diverticulum is a true diverticulum typically located on the antimesenteric border, is the result of a failure in the obliteration of the vitelline duct (omphalomesenteric duct), which connects the yolk sac to the midgut through the umbilical cord [1,3,14]. MD contains all four layers of the intestinal wall with its independent blood supply [3,10,15]. The persistence of the vitelline duct may lead to the formation of a Meckel's diverticulum, a fistula, an umbilical sinus, a fibrous cord, an enterocystoma, a mesodiverticular band, and a congenital umbilical hernia [3,14].

In adults the most frequent location of the Meckel's diverticulum is on the antimesenteric border of the terminal ileum, 60–100 cm from the Bahuinis valve; in children, the usual place is on the antimesenteric side of the terminal ileum, 30–60 cm from the Bahuinis valve [9,14,16]. The 80–90% of the MD are asymptomatic and equally found in males and females; the complications are three to four times higher in men [3,6,17], been the most common an intestinal obstruction, hemorrhage, diverticulitis, ulceration, and perforation [3,4,14,17]. A heterotopic tissue is found in the diverticula in approximately 50% of the patients (gastric, duodenal, colonic, and pancreatic are the most frequent) [3,4,14,17]. Park JJ et al. [17] performed a review of 1476 patients with MD; they divided the patients into two groups, one symptomatic and the other asymptomatic. Only 16% of the patients were symptomatic with a mean age of 31 ± 23.6 years; the male to female ratio was 3:1, and the clinical presentation was bleeding in 38%, obstruction in 34%, and diverticulitis in 28%. They found that the surgical remove of an asymptomatic MD had a morbidity of 20%, and a mortality of 3%, much higher than the symptomatic group (13% of morbidity, and 0% of mortality).

In the context of the MD, the intestinal obstruction may be due to intussusception, adhesive band, volvulus, herniation (as in the case presentation), and enterolith formation [3,14,17]. A hernia containing a Meckel's diverticulum is called Littre's hernia and is

Table 1
Literature review.

Author and year	Number of patients	Surgery performed
Losanoff JE and Kjossev KT. 2000 [8]	1	Not found
Urakawa M et al. 2009 [7]	1	Not found
Blando-Ramírez JS et al. 2014 [9]	1	Intestinal resection with anastomoses
Tas I et al. 2015 [4]	1	Intestinal resection with anastomoses
Fajardo R et al 2011 [10]	1	Intestinal resection with anastomoses
Emre A et al. 2013 [6]	1	Diverticulectomy
Luna-Lugo G and Guzman-Sanchez C. 2011 [5]	1	Intestinal resection with anastomoses
Albu E et al. 1992 [11]	1	Not found
Mazza L et al. 2006 [18]	1	Diverticulectomy
Tauro LF et al. 2009 [19]	1	Not found
Yang JG et al. 2008 [12]	Not found	Not found

detected in the inguinal region in 50%, in the femoral region in 20%, the umbilical region in 20%, and other locations in 10% [1,2,9,16]. Is not unusual to find a strangulated Littre's hernia, but is exceedingly rare to see a duplication of a Meckel's diverticulum, with just a few cases reported in the literature (See Table 1). We could not find another publication of a simultaneous strangulated Littre's hernia with MD duplication as in our case.

The management of the MD will depend on the clinical presentation. When the Meckel's diverticulum is symptomatic, the surgical resection is required, but when it is asymptomatic, there is not a consensus on the best treatment. In 1976, Soltero and Bill [20], concluded that 800 incidental diverticulectomies were needed to prevent one death, with a postoperative morbidity of approximately 10%; therefore, they recommended against the removal of an incidental MD.

The symptomatic MD and the Littre's hernia require surgical resection [1,17]; both, diverticulectomy and intestinal resection with primary anastomoses are surgical options [1,3,4,14]. It is recommended to do an intestinal resection with primary anastomoses when there is edema or inflammation at the base of the diverticulum [1], there is palpable ectopic tissue at the diverticular-intestinal junction, intestinal ischemia or perforation [3,17]. Park JJ et al. [17] showed that the patient age younger than 50-years, male sex, a diverticulum length greater than 2 cm, and an ectopic or abnormal features within a diverticulum were all risk factors associated with symptomatic diverticula; with an incidence of 17% when 1 criterion was met, 25%, 42%, or 70%, when two, three or all of the criteria were met, respectively. We did only a diverticulectomy of the MD that was in the Littre's hernia as recommended [1]; the decision to perform this particular technique was because the small bowel incarcerated regained its vascular flow with normalization of the color, and the diverticulum has not a palpable mass. The second Meckel's diverticulum did not accomplish any of the Park criteria and was left in site.

4. Conclusion

The Meckel's diverticulum is a rare entity, been the most common anomaly of the gastrointestinal tract. The management of the symptomatic MD is surgery, with a weak recommendation about the preferred technique; the asymptomatic Meckel's diverticulum could be resected if accomplishes the Park criteria [17]. When dealing with a Littre's hernia, the MD should always be removed, an intestinal resection with primary anastomoses should be attempt if the patient presents a palpable ectopic tissue at the diverticular-intestinal junction, intestinal ischemia or perforation. As to our knowledge, this is the first case of an incarcerated Littre's hernia with duplication of a Meckel's diverticulum.

Conflicts of interest

All authors declare no conflict of interest about the publication of this article.

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Ethical approval

The written consent was sign by the patient.

Consent

No personal information is given nor modified.

Author contributions

López-Lizárraga CR: study concept, writing the paper, final decision to publish, data collection.

Sánchez-Muñoz MP: study concept, data collection, writing the paper, final decision to publish.

Juárez-López GE: data collection and analysis.

Pelayo-Orozco L: data collection and analysis.

De la Cerda-Trujillo LF: data collection, writing and analysis.

Ploneda-Valencia CF: writing the paper, data collection, final decision to publish.

Registration of research studies

Is a case report.

Guarantors

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