Littré's hernia – a case of a Meckel's diverticulum in a strangulated femoral hernia and literature review

Hérnia de Littré – a propósito de um caso clínico de um divertículo de Meckel numa hérnia femoral estrangulada

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RESUMO

Introdução: O divertículo de Meckel é a anomalia gastrointestinal congénita mais comum e resulta da obliteração incompleta do ducto vitelino. A hérnia de Littré define-se pela presença de um divertículo de Meckel no saco herniário. O estrangulamento do divertículo de Meckel numa hérnia femoral é uma entidade extremamente rara que requer correção cirúrgica imediata. **Caso Clínico:** Reportamos o caso clínico de uma mulher de 78 anos que recorreu ao serviço de urgência por uma hérnia femoral dolorosa e encarcerada à esquerda, sem sinais ou sintomas de obstrução intestinal associados. Durante a intervenção cirúrgica, aquando da abertura do saco herniário, foi identificado um divertículo de Meckel, procedendo-se então a diverticulectomia. A ansa de intestino delgado não se encontrava herniada nem tinha sinais de isquemia, pelo que não foi necessário realizar uma enterectomia. O defeito herniário foi subsequentemente corrigido através da técnica de Lichtenstein modificada. **Discussão:** A sintomatologia de um divertículo de Meckel complicado é inespecífica e dependente do tipo de complicação. O diagnóstico de hérnia de Littré é exigente e difícil de estabelecer durante o pré-operatório, uma vez que a sua apresentação clínica é ambígua e a imagiologia tem um papel limitado. Quando a obstrução do intestino delgado é provocada por um divertículo de Meckel complicado, a intervenção cirúrgica tem como objetivo principal remover o divertículo e corrigir a patologia associada, independentemente da abordagem utilizada ser laparoscópica ou cirurgia aberta. **Conclusão:** No caso de uma hérnia de Littré estrangulada o tratamento é baseado na condição clínica do doente e na experiência do cirurgião. Não obstante, o tratamento requer, pelo menos, uma ressecção completa do divertículo para evitar complicações futuras.

Palavras Chave: *hernia de Littré, divertículo de Meckel, hérnia femoral, hérnia estrangulada.*

ABSTRACT

Introduction: Meckel's diverticulum is the most common gastrointestinal congenital anomaly and it results from an incomplete obliteration of the vitelline duct. Littré's hernia is defined by the presence of a Meckel's diverticulum in a hernia sac. A strangulated Meckel's diverticulum in a femoral hernia is an extremely rare entity and its management requires prompt surgery. **Case Report:** We describe a case of a 78-year-old woman that presented with a painful incarcerated femoral hernia in her left groin. She did not have any signs or symptoms of bowel obstruction. During surgery, a Meckel's diverticulum was identified in the hernia sac. The

small bowel was not herniated and did not show signs of ischaemia, thus, an enterectomy was not required. A diverticulectomy was performed and the hernia defect was subsequently repaired with a modified Lichtenstein technique. **Discussion:** The symptomatology of a complicated Meckel's diverticulum is non-specific and dependent on the type of complication. The diagnosis of Littré's hernia is challenging and difficult to establish preoperatively since its presentation is ambiguous and imaging has a limited role. When small bowel obstruction is caused by a complicated Meckel's diverticulum, the main aim of surgery is to remove the diverticulum and to correct the associated pathology, either by laparoscopy or by an open surgical approach. **Conclusion:** A patient's condition and surgeon based management is generally adopted in case of a strangulated Littré's hernia. Nevertheless, it requires complete resection of the diverticulum to avoid future complications.

Keywords: *Littré's hernia, Meckel's diverticulum, femoral hernia, hernia strangulation.*

INTRODUCTION

Meckel's diverticulum (MD) is the most frequent gastrointestinal congenital anomaly[1]. It was first described by Fabricus Hildanus in the sixteenth century, although it bears the name of Hohann Friedrieck Meckel, who described it in detail and predicted its important clinical implications in 1809[2,3,4]. These clinical implications were suspected long before clinical appreciation of inflammation of the appendix[4]. The first successful diverticulectomy was conducted by Oderfeld in 1892[4].

MD is the remnant of the prenatal yolk stalk. Until it normally regresses between the sixth and ninth weeks of fetal life, the yolk stalk (also called omphalomesenteric duct) connects the primitive gut and the yolk sac of the developing embryo. When the process of regression fails, a MD may occur[5,6].

MD characteristics follow the "rule of 2's", since it occurs in about 2% of the population, 2% of which are symptomatic and it is usually 2 inch length (5 cm) and located 2 feet (60 cm) from the ileocecal valve[6]. Several complications of MD have been described, such as obstruction, diverticulitis, hemorrhage and malignant degeneration. Its presence in a hernia sac is a very uncommon complication[7].

Femoral hernia is the second most common hernia of the groin area. It most commonly presents as a flexible, round, dome-shaped, walnut-sized bulge on the medial side of the thigh, 2-3 cm below the inguinal ligament. It is more common in women than men and reaches up to 34% of all hernias occurring in women. It occurs in less than 5% of hernia cases in men. Due to the rigid and narrow characteristics of the femoral canal, incarceration and strangulation of the hernia contents is highly prevalent, reaching as much as 60%. Thus, prompt surgery is mandatory to avoid complications[3].

Alexis Littré, a French surgeon, in 1700s first described incarcerated femoral hernias containing a MD[8,9]. Littré's hernia (LH) is defined by the presence of a MD in any kind of hernia sac [4,7]. LH is extremely rare and about 50 cases have been reported since it was first described[8,10]. Strangulation of a MD in a femoral hernia is even rarer[3,10].

We describe a case of a 78-year-old woman with an incarcerated femoral hernia containing a MD, which was identified during surgery.

CASE REPORT

A 78-year-old caucasian woman, with no significant past medical history, attended our emergency department with complaints of a non-reducible painful mass in her left groin which had been present during the course of two weeks. She denied nausea, vomiting, abdominal pain and altered bowel movements, thus no signs of bowel obstruction were present. She denied the presence of fever at any time, as well as night sweats and weight loss. There was no history of trauma nor of previous abdominal surgeries.

Physical examination was unremarkable except for a non-pulsatile, non-reducible mass, about 3 cm in diameter, located in the left groin. The mass was tender at palpation, hard in consistency and the overlying skin was erythematous. She did not present abdominal pain or fever. Strangulated groin hernia was diagnosed clinically and the patient prepared for urgent surgical intervention. Additionally, blood was collected and laboratory tests conducted: all parameters were within our laboratory ranges, except for a C-reactive protein of 2.01 mg/dL.

Under general anesthesia, a left inguinal incision was made and a strangulated femoral hernia identified. During surgery, upon opening the femoral hernia sac, a thick hemorrhagic non-ruptured MD was identified on the antimesenteric side of the small bowel (figure 1). The small bowel loop was not herniated and did not show signs of ischemia, thus, an enterectomy was not necessary. A diverticulectomy was performed with a linear staple GIA® 60. There was no spillage of gastrointestinal contents thus a modified Lichtenstein technique was adopted to correct the hernia defect. This was done by, after handling the sac with a transfixing suture, exposing Cooper's ligament so that a triangular shaped extension of the propylen mesh could be secured to it and to the inguinal ligament with a continuous Prolene® suture. Interrupted sutures were used to secure the medial and upper margins of the mesh. Wound closure was performed in a standard manner.

A seroma of the surgical wound complicated the postoperative period and it was managed with drainage and regular dressings. The patient was discharged on the eighth postoperative day.

Histology report described a saccular structure measuring 4 cm in length and 2,5 cm in width with small bowel wall characteristics. No ectopic mucosa or signs of malignancy were observed. The resected structure was identified as a MD, thus confirming the diagnosis of LH.

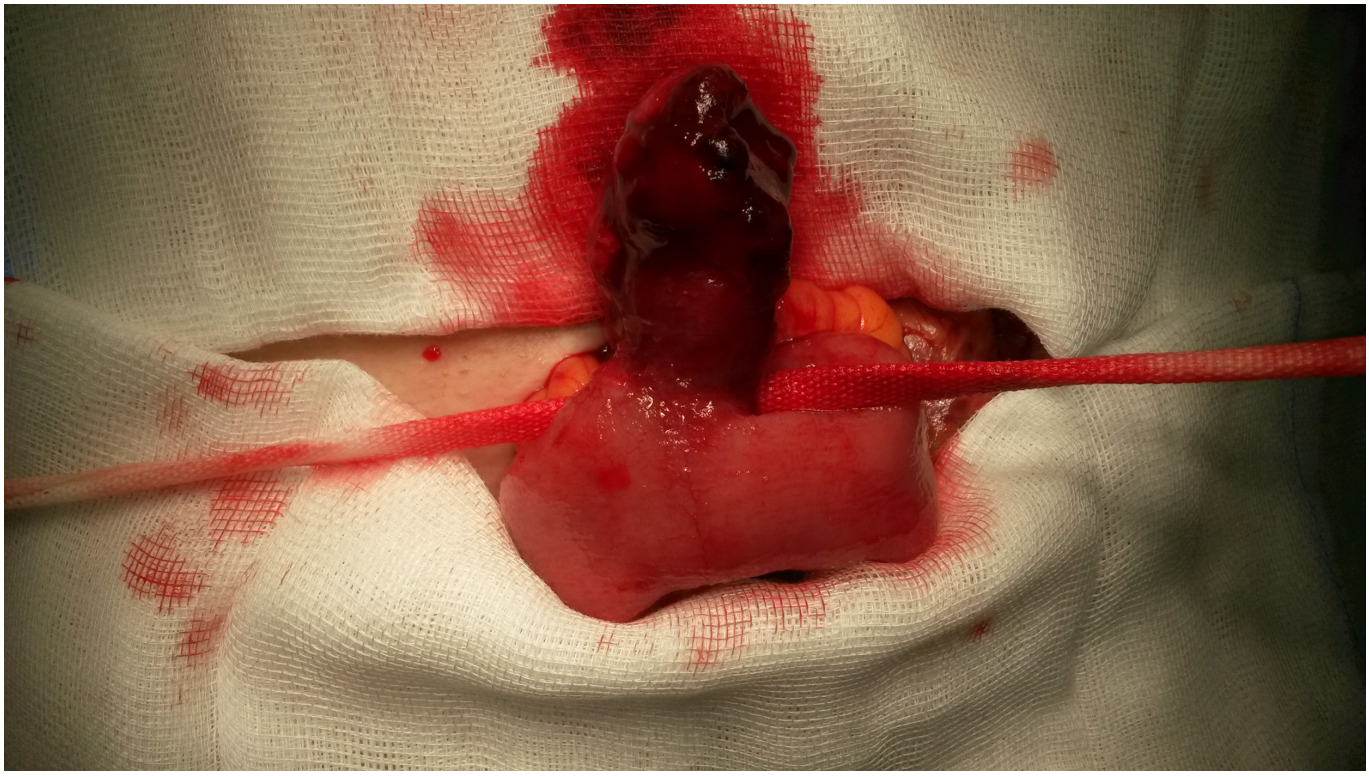

Fig. 1 – **Meckel's diverticulum with inflammatory signs.**

DISCUSSION

The embryologic origin of MD relies on the development of the midgut. The primitive yolk sac divides into a larger portion, which constitutes the primitive gut, and into a smaller one, which continues as a yolk sac near the placenta. These two portions remain connected by the omphalomesenteric duct, lying within the umbilical cord. Most commonly, this duct begins to obliterate between the sixth and the ninth week of gestation. When obliteration is incomplete and the proximal end of the vitelline duct persists, a MD is formed[4]. Other anomalies that may result during the obliteration process are umbilical-intestinal fistula, umbilical *sinus*, fibrous cords attaching the distal ileum to the abdominal wall, enterocystoma and mesodiverticular band[2,5,6]. An association of a variety of pathologic conditions with MD has been found. These conditions are intestinal obstruction due to band, volvulus, intussusception, regional enteritis, herniation, enterolith formation, diverticulitis, foreign bodies, fistula, and tumors such as angioma, lipoma, leiomyoma, fibroma, carcinoid, adenocarcinoma and sarcoma. Anorectal malformations, exomphalos, central nervous system and cardiovascular malformations, esophageal atresia and angiodysplasia are other associated pathological conditions [2].

MD can be found on the antimesenteric border of the terminal ileum, 45 cm – 90 cm proximally to the ileocecal valve[2,5]. The diverticulum may be long and narrow or present as a short outpouch of the ileum[4]. More commonly, and as we can see in our case in (figure 1), the tip of the diverticulum is unattached, although it can be adherent to adjacent bowel or mesentery or connected to the undersurface of the umbilicus by fibrous remnants of the obliterated duct[4]. MD duplication has been reported[1].

MD is a true diverticulum as it contains all the three layers of the intestinal wall[2,8]. It has its own blood supply from the superior mesenteric artery and it is vulnerable to infection and obstruction[2,5]. As a result of pluripotency of vitelline duct cell lining, heterotopic mucosa may also be found[5]. About half of MD contain ectopic gastric or pancreatic mucosa. Less commonly, jejunal, duodenal, colonic mucosa, endometriosis and hepatobiliary tissue may be present[2,5]. In our patient's pathology results only small bowel mucosa was found.

The range of incidence of MD varies amongst the literature from 0.2% to 4% of the population. Although autopsy studies report an incidence of 0,3%, surgical studies more commonly refer to a 2% incidence[2,3,4,5,6,8,9]. MD is in most instances an incidental finding during a small bowel contrast study, during a laparoscopy or a laparotomy done for an unrelated condition[4,5]. It is as common in males as it is in females, although the rate of complications is higher in men[2,5]. The ratio of complications vary from 1.8:1 to 3:1[5]. No explanation in the literature for this fact was found.

Although clinical manifestations are more common in pediatric age, MD can cause symptoms at any age. The presence of a MD carries a 4% to 6% lifetime risk of developing a complication[5]. Male gender, age (< 10 years), the length of the diverticulum (> 4 cm) and the presence of gastric or pancreatic heterotopic tissue in the diverticulum have been pointed out as risk factors for complications[7].

The symptomatology of a complicated MD is non-specific and dependent on the type of complication. Hence the diagnosis is difficult to establish. MD may present itself as an anaemic syndrome, features of appendicitis, bowel obstruction, acute peritonitis or as haemorrhagic shock[6].

Gastrointestinal bleeding due to ulceration is the most common complication of MD in the pediatric age group, specifically in children aged 2 years or younger[5,8]. The ectopic gastric mucosa and the adjacent ileal mucosa are vulnerable to ulceration, which may cause gastrointestinal bleeding[2,5,6]. In these patients, regardless their age group, segmental resection is indicated since bleeding from the adjacent ileum may recur postoperatively if only a simple diverticulectomy is performed[2,5]. In the adult population, bowel obstruction is the most common complication and it is usually followed by inflammation and bleeding[5,8]. In the case of obstruction the presentation is similar no

matter its cause. Signs and symptoms of small bowel obstruction, such as absolute constipation, spasmodic abdominal pain and vomiting, are typically present [5]. The mechanisms by which a MD can cause intestinal obstruction are: volvulus of small intestine around a fibrous band extending from MD to umbilicus; intussusception in which the MD sags into the bowel lumen and serves as a lead point to allow telescoping of the small intestine; mesodiverticular band known by entrapment of small bowel beneath the diverticulum blood supply; chronic diverticulitis causing stricture; MD lithiasis and LH[5]. When small bowel obstruction is caused by a complicated MD, the main aim of surgery is to remove the MD and to correct the associated pathology. This can be performed under a laparoscopic or an open surgical approach[5].

Femoral hernias are characterized by the protrusion of abdominal contents through the femoral ring. They are more frequent in women than in men in a 4:1 ratio and constitute about one third of all hernias in women[3]. A femoral hernia may present as a small bulge of approximately 5 cm, dome shaped, in the medial aspect of the thigh, located 2 cm to 3 cm below the inguinal ligament. The right side is more commonly affected[3]. Since the femoral canal is narrow and rigid, the contents incarcerate or strangulate in up to 60% of cases[3,8]. In most cases, the lacunar ligament is responsible for the constriction[3].

Femoral hernia is always acquired. It may contain, amongst others, stomach, omentum, colon, small bowel, the appendix, urinary bladder or a fallopian tube[3].

LH is the type of herniation where a MD is the content of the hernia sac. Although Littré first described it as a femoral hernia, half of Littré's hernias are inguinal. It may also occur as an umbilical hernia[1,8,11,10]. It is more common in women and in the right inguinal canal[7,9,10].

Clinically, there are no specific signs to recognize the extremely rare cases of incarcerated LH preoperatively[7,8]. When only the diverticulum is located within the hernia sac the bowel lumen is not obstructed[8]. Hence, the signs and symptoms of an incarcerated MD on presentation are thought to progress slower than a hernia involving the complete lumen of the small bowel[9,11]. If systemic inflammation with pain and fever is present, it might be less severe and occur late in the course of the disease[8,9]. Incarceration of a hernia is a term used to describe entrapment of the hernia contents. It leads to swelling of the trapped tissue. Ultimately, the entrapment and the swelling reduce arterial flow, which results in ischemia and necrosis of the hernia contents – strangulation[8]. Femoral hernia incarceration is a potentially life-threatening complication. Although it usually presents as a non-reducible mass, on the medial aspect of the thigh, other clinical symptoms of an incarcerated femoral hernia are often ambiguous and they greatly depend on the content of the hernia sac. Since a MD, an appendix, a fallopian tube or bladder within the hernia sac may not present with gastrointestinal occlusion symptoms, the correct diagnosis often occurs during surgery[3]. On the contrary, there might be associated symptoms of bowel obstruction if, for example, the bowel becomes kinked when it is caught inside the femoral ring[9]. Skin changes, abdominal distension or even peritonitis may accompany this presentation[8,9]. In our case, considering the relatively long history of symptoms and the absence of gastrointestinal tract complaints, a total intestinal obstruction was not likely.

The early diagnosis of groin hernia may be even more complicated due to a broad spectrum of differentials. Great saphenous vein varicosity, femoral artery aneurysm, ectopic testis, psoas abscess, cystic masses and enlarged lymph nodes may present as a groin mass[3,8].

Being widely available, non-invasive and cheap, ultrasonography might be useful in the diagnosis of LH. By demonstrating characteristics of intestine wall, ultrasonography can help excluding other differential diagnosis thus avoiding unnecessary invasive interventions such as biopsy and groin exploration[8]. Misiak et al. refer to ultrasonography as a contributor to a correct preoperative diagnosis[3]. On the contrary, Magagi et al.[7] stated that ultrasonography does not offer further information indicating the presence of a Littré's hernia while computed tomographic scan could

demonstrate MD as a blind-ending tubular formation communicating with the distal ileum[7].

Strategies on MD complications management have been based on the patient's condition and on the surgeon's perspective, which seems to be appropriate in an emergency condition[5].

There is no consensus on the best treatment for an asymptomatic MD. Its surgical removal may comprise an increased risk of morbidity and mortality when compared to symptomatic MD. López-Lizárraga et al.[1] suggested an asymptomatic MD should be resected if it follows the Park criteria: patient younger than 50 years, male sex, diverticulum length greater than 2 cm and an ectopic or abnormal features within the diverticulum[1].

The treatment of LH is surgical. It includes resection of the diverticulum and hernia defect repair[5]. Resection of MD is recommended due to the risk of complications[7]. MD surgical resection may be done by simple diverticulectomy using a linear GI stapler or by segmental resection of the involved small bowel and primary anastomosis[11]. Some authors prefer a wedge resection of the diverticulum, justifying their choice by the lack of studies comparing the outcome of wedge resection as against resection and anastomosis[1,7]. Nevertheless, in case of perforation, bowel ischaemia, edema or inflammation at the base of the diverticulum, or when there is palpable ectopic tissue at the diverticular-intestinal junction, a resection and small bowel anastomosis is recommended[1,11].

If an open repair approach is adopted, traditional methods to repair the hernia defect should be undertaken after MD resection[11]. The repair is generally not complicated by MD removal. However, a theoretical increased risk of hernia site infection has been pointed out[11]. An incision above the inguinal ligament for femoral hernia defect correction is recommended for younger patients. Although femoral access tends to cause less perioperative trauma, it has a higher percentage of recurrence and makes the resection of the ischaemic intestinal fragment highly difficult to conduct. Nevertheless, Misiak et al.[3] recommend femoral access as the first-choice approach for elderly patients. An incision above the inguinal ligament seemed more appropriate in our patient as she was very active in her daily routine. This approach allowed us to examine the bowel without difficulty. The ileum was not incarcerated and did not show any signs of ischaemia nor alteration upon palpation of the diverticular-intestinal junction, thus only a diverticulectomy was performed.

The advent of laparoscopic surgery has altered the management of all abdominal hernias, including LH. It is a safe, inexpensive and efficient method for the diagnosis and treatment of LH[10]. Chan et al.[12] suggest that the limited preperitoneal dissection, without dissection and exploration of the edematous groin structures, theoretically, makes laparoscopic repair superior to traditional open repair in the management of incarcerated LH. The MD can be excised by endoscopic stapler or via an extended umbilical wound. The excision via an extended umbilical wound, in pediatric reports, has been the prefered method as it allows direct vision and palpation of ectopic mucosa in the adjacent ileum when present, thus leading to a small bowel resection[12].

CONCLUSION

The incidence of a MD in a femoral hernia is extremely rare. LH is difficult to diagnose preoperatively, as the clinical symptoms are not specific. A patient's condition and surgeon based management is generally adopted to this emergent situation. Ultrasound might be an useful diagnostic tool. The appropriate treatment of LH requires, at minimum, a complete resection of the diverticulum to avoid future complications. A laparoscopic approach to this type of hernia has been shown to be safe and effective and shows additional advantages in comparison to an open repair.

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