CASE REPORT

Mixed Littre's Hernia with Concomitant Ipsilateral Cryptorchidism in an Infant: A Triple Surgical Challenge

Hernia de Littre Mixta con criptorquidia ipsilateral concomitante en un lactante: Un triple desafío quirúrgico

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ABSTRACT

Background: Littre's hernia, defined by the presence of a Meckel's diverticulum within a hernia sac, is an exceptionally rare condition in pediatric surgery. Due to the lack of specific external signs and its often misleading acute presentation, it is most often identified intraoperatively. **Case presentation:** A 40-day-old male infant presented with irritability, intermittent abdominal discomfort, vomiting, right-sided scrotal swelling, and refusal to feed. Physical examination revealed an empty right hemiscrotum and tenderness in the inguinal region. The initial diagnosis suggested torsion of an undescended testis. Surgical exploration revealed an incarcerated Meckel's diverticulum within the right inguinal hernia sac showing signs of ischemia. A segmental small bowel resection with end-to-end anastomosis was performed, followed by hernia repair using the Bassini technique. **Conclusion:** Littre's hernia is an uncommon finding in infancy and may mimic other acute scrotal or inguinal pathologies. Early recognition and prompt surgical intervention are crucial for achieving favorable outcomes.

Keywords: Littre hernia; Inguinal hernia; Infant; Bowel resection; Meckel diverticulum

RESUMEN

Introducción: La hernia de Littre, definida por la presencia de un divertículo de Meckel dentro de un saco herniario, es una entidad excepcionalmente rara en la cirugía pediátrica. Debido a que el divertículo no produce signos externos específicos y su presentación aguda puede ser engañosa, generalmente se identifica durante el acto quirúrgico. **Presentación del caso:** Lactante masculino de 40 días de vida con irritabilidad, molestias abdominales intermitentes, vómitos, aumento de volumen escrotal derecho y rechazo al alimento. El examen físico reveló un hemioscroto derecho vacío y dolor a la palpación en la región inguinal. El diagnóstico inicial sugirió una torsión de testículo no descendido. La exploración quirúrgica evidenció un divertículo de Meckel incarcerado dentro del saco herniario inguinal derecho con signos de isquemia. Se realizó resección segmentaria del intestino delgado con anastomosis término-terminal y reparación de la hernia mediante la técnica de Bassini. **Conclusión:** La hernia de Littre es un hallazgo infrecuente en la infancia que puede simular otras patologías escrotales o inguinales agudas. El reconocimiento temprano y la intervención quirúrgica oportuna son claves para lograr resultados favorables.

Palabras clave: Hernia de Littre; Hernia inguinal; Lactante; Resección intestinal; Divertículo de Meckel

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INTRODUCTION

Littre's hernia, first described in the early 18th century, is defined by the presence of a Meckel's diverticulum within a hernia sac.[1] Meckel's diverticulum is the most common congenital anomaly of the gastrointestinal tract, resulting from the incomplete obliteration of the omphalomesenteric duct. It affects approximately 2% of the general population and is typically asymptomatic.[2] The diverticulum may contain heterotopic mucosa, most commonly gastric (60-80%), but also pancreatic or colonic tissue. Other ectopic tissues, such as duodenal or endometrial tissue, have been reported, which predispose the patient to complications such as bleeding, ulceration, or perforation. These complications underscore the need for careful assessment when identified.[2,3]

Inguinal hernias represent the most frequent site for Littre's hernia, with the right side being more commonly affected than the left, followed by femoral and umbilical hernias.[4] Clinically, it often presents as a tender, irreducible groin mass indistinguishable from other types of incarcerated hernias. Preoperative diagnosis is rare, as imaging modalities often show bowel loops but are not specific for Meckel's diverticulum.[4,5] Consequently, the diagnosis is usually made intraoperatively during the management of complications such as obstruction or ischemia. Standard repair involves resection of the diverticulum and hernia repair using either an open or laparoscopic approach, depending on the presentation and surgeon expertise, to avoid morbidity associated with strangulated bowel.[4,5]

An unusual case of incarcerated Littre's hernia is reported herein, illustrating its capacity to mimic testicular torsion in neonates and infants and reinforcing the necessity of early surgical exploration.

CASE REPORT

A 40-day-old term male infant, born at 41.3 weeks via vaginal delivery with a birth weight of 3250 grams and Apgar scores of 9/9, exclusively breastfed, was brought to the hospital due to irritability lasting 10 hours. According to the mother, the infant began exhibiting signs of discomfort, intermittent crying without relief, and worsening abdominal pain, accompanied by vomiting.

On physical examination, the infant appeared restless and ill. Axillary temperature was 36.8°C. The abdomen was soft, no masses. Examination of the groin revealed swelling at the right inguinal region extending to the scrotum. The area was very tender, firm, and thickened. The right hemiscrotum was empty, while the left testis was palpable in the scrotum. The initial diagnosis was torsion of an undescended testis.

Preoperative workup

Laboratory results showed hemoglobin at 112 g/L, blood type O-, and serum electrolyte analysis indicated mild hyponatremic dehydration. Fluid and electrolyte imbalances were corrected, and a single dose of cefazolin was administered for prophylaxis.

Surgical procedure

A transverse inguinal incision (~3 cm) was made along the inguinal crease. Dissection proceeded through the layers to the external oblique aponeurosis. Upon opening the inguinal canal, an incarcerated loop was identified within the hernia sac. The right testis was compressed by the hernia and showed signs of ischemia, which improved after it was wrapped in gauze soaked in warm saline. An incarcerated Meckel's diverticulum (~2.5 cm in length) with ischemic changes was revealed after opening the hernia sac (Fig.1). A segmental small bowel resection was performed, including the diverticulum and adjacent ileum (approximately 10 cm), followed by an end-to-end anastomosis (Fig. 2).

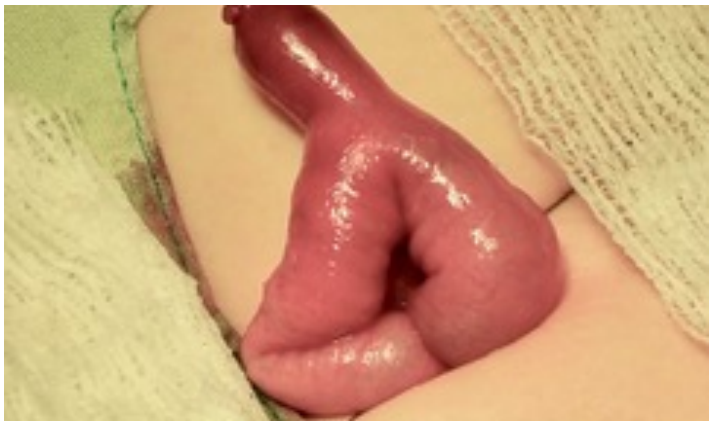

Figure 1. A segment of small bowel (black star) containing a Meckel's diverticulum (pointed by the blue arrow) delivered through the inguinotomy, showing discoloration throughout its entire length.

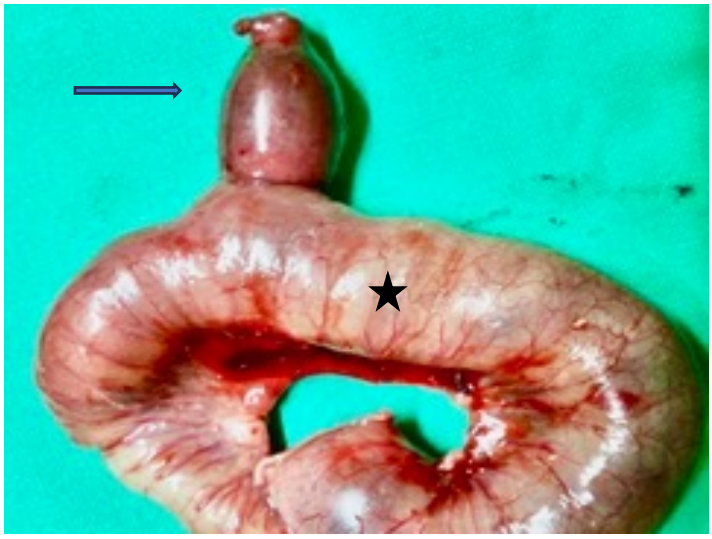

Figure 2. Resected segment of small bowel (black star) containing the Meckel's diverticulum (pointed by the blue arrow).

Hernia repair was completed with high ligation of the hernia sac. Given that the testis appeared viable, a dartos pouch orchiopexy was performed. The posterior wall was reinforced using the Bassini technique.

The patient tolerated the procedure well and was discharged on postoperative day eight. Pathology confirmed a Meckel's diverticulum with ischemic necrosis and inflammatory foci. At the 6-month follow-up, there was no evidence of recurrence or testicular atrophy and the wound was healed without complications (Fig.3).

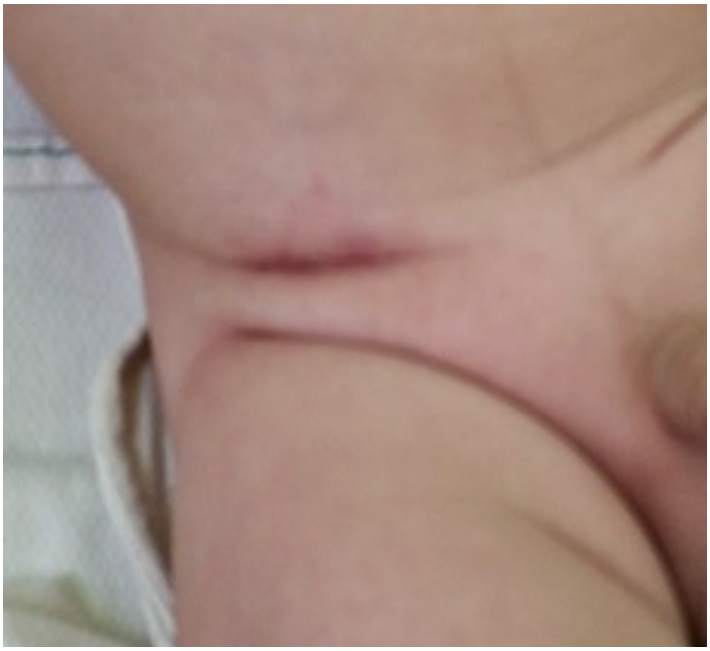

Figure 3. Inguinal area and surgical wound, two weeks after surgery.

DISCUSSION

Littre's hernia was first described in 1700 by the French surgeon Alexis Littre, who reported what he termed a "new kind of hernia" containing a small, bladder-like protrusion of bowel, this structure was later recognized as Meckel's diverticulum, documented in the *Histoires de l'Académie Royale des Sciences*.[1] More than a century later, in 1809, Johann Friedrich Meckel the Younger, published the first comprehensive anatomical and embryologic description of this ileal diverticulum in the *Archiv für die Physiologie*, defined as a congenital remnant of the omphalomesenteric duct.[6]

Meckel's diverticulum affects approximately 2% of the general population and is classically summarized by the "Rule of Twos." This mnemonic describes its epidemiological and anatomical characteristics: it is most often diagnosed before two years of age; it typically measures about two inches (5 cm) in length and 2 cm in diameter; it is located roughly two feet (60 cm) proximal to the ileocecal valve; it demonstrates a 2:1 male predominance; and it becomes clinically symptomatic in approximately 2% of patients.[2,3,7] In their large review of 1,476 cases, Park *et al.*[8] demonstrated that the predominant symptomatic presentation differed by age group: intestinal obstruction prevailed among children, while adults more frequently manifested with gastrointestinal bleeding.

Inguinal hernias are themselves common in male infants, with an incidence of 1% to 5%, and substantially higher rates observed in preterm neonates. In stark contrast, Littre's hernia, defined by the incidental presence of a Meckel's diverticulum within the hernial sac, is exceedingly rare in infancy. It has been reported predominantly as isolated pediatric case reports, particularly in children younger than one year.[8]

Anatomically, the inguinal canal is the most frequent site for Littre's hernia, accounting for approximately 50% of cases. This is followed by femoral hernias (20%) and umbilical hernias.[7] Less common locations include obturator, Spigelian, and ventral hernias. Notably, the anatomical spectrum was recently expanded in 2023 when Trigui *et al.*[9] reported the first documented case of Littre's hernia arising in an incisional site.

The preoperative diagnosis of Littre's hernia is unequivocally one of the most demanding aspects of its management, primarily because its presentation often mimics other acute inguinal emergencies, such as testicular torsion, simple incarcerated hernia, or even appendiceal herniation (Amyand's hernia).[4,10] This diagnostic difficulty is exacerbated by the notorious limitations of modern imaging modalities. Current evidence indicates that despite the availability of techniques like ultrasonography, computed tomography, scintigraphy, or technetium-based scans, preoperative confirmation is rarely possible even when multiple methods are used in combination. [4,5,7,11,12]

The condition's nonspecific clinical manifestations and the lack of pathognomonic radiologic features, explain why most cases are identified incidentally during operative exploration.[11,12] While the absence of pathognomonic physical signs leaves the surgeon dependent on subtle clues, a careful history in older patients can provide essential hints: episodes of rectal bleeding, chronic or intermittent right iliac fossa pain, or a history of difficult hernia reduction should raise suspicion of an incarcerated Meckel's diverticulum.[4,8,9] In our young patient, however, the association of irritability, vomiting, mild inguinal swelling, and an empty right hemiscrotum reasonably suggested torsion of an undescended testis, illustrating the significant overlap between Littre's hernia and other time-sensitive pediatric groin pathologies. Ultimately, this persistent diagnostic ambiguity and the risk of intestinal compromise justify prompt surgical exploration whenever clinical deterioration or persistent, unresolving symptoms are present, even without definitive imaging evidence. Moreover,

this case highlights the importance of clinical judgment in resource-limited settings, where advanced imaging modalities may not be readily available and timely surgical exploration remains the cornerstone of management in acute pediatric groin emergencies.

Littre's hernia is classified based on intraoperative findings as either true or mixed. The true Littre's hernia is the more commonly reported subtype, defined by the presence of an isolated Meckel's diverticulum within the hernia sac. In contrast, the mixed Littre's hernia is less frequently described and involves the diverticulum alongside an adjacent segment of the small intestine.[11,12]

In our patient, the hernia corresponded to the mixed type. The sac contained the Meckel's diverticulum along with approximately 5-7 cm of ileal loops on either side of the diverticulum, all displaying early changes in coloration consistent with compromised viability. This mixed presentation, which carries a higher risk of intestinal obstruction and strangulation compared to the true type, warrants careful consideration. For instance, Odongo *et al.*[12] in 2023 described a rare presentation of the mixed subtype featuring a perforated, yet asymptomatic, Meckel's diverticulum, further highlighting the variability in clinical and surgical pathology associated with this classification.

The management of Littre's hernia in the pediatric population necessitates a two-step approach: addressing the Meckel's diverticulum and repairing the hernia defect.[3,12] In specific scenarios, a third procedure, such as orchiopexy, is required to correct coexisting undescended testes, as in the case presented.

Surgical intervention for the Meckel's diverticulum is mandatory when symptomatic or complicated by obstruction, inflammation, or perforation. The preferred strategy is either a wedge resection or segmental small-bowel resection, determined by the extent of involvement and the viability of the adjacent ileum.[3,4,8,13] The decision regarding the extent of resection is crucial and often hinges on the presence of heterotopic tissue at the diverticular base, which must be completely removed to minimize long-term risks.[8] In instances where this tissue is suspected (e.g., via intraoperative palpation), a segmental intestinal resection with primary end-to-end anastomosis is the appropriate surgical choice, as was performed in our patient.[12,14]

For asymptomatic, incidentally discovered Meckel's diverticulum in children, there remains no definitive consensus. To guide this decision, the Robijn *et al.*[15] risk-scoring system provides a structured approach. This system incorporates factors such as patient sex, age, diverticular length, and the presence of a fibrous band, recommending prophylactic resection when the total score suggests a significant future complication risk.

The standard of care for the hernia defect repair in pediatric patients is primary tissue repair (herniorrhaphy). The use of prosthetic mesh is systematically avoided in children due to concerns regarding interference with growth and a lack of long-term safety data in this age group. Furthermore, its use is strictly limited in the setting of contamination, such as a perforated Meckel's diverticulum or compromised bowel.[15] Techniques like the Bassini technique for posterior wall reinforcement may be employed when required by tissue weakness, a measure described in complex pediatric cases.[16]

In selected, non-obstructed patients, a combined or hybrid approach, utilizing open repair for the defect and laparoscopy for assessing bowel viability or facilitating Meckel's diverticulum resection, can be leveraged, offering the benefits of minimally invasive exploration without compromising the safety of the standard open repair.[5,16,17]

Due to the presence of a viable right undescended testis, the third and final step of the procedure was a right orchidopexy. To the best of our knowledge, the association of a Littre's hernia with this specific urological condition represents the first reported case in the literature.

Postoperative outcomes in the majority of reported patients, including our own, have been favorable. However, a systematic review by Răcăreanu *et al.*[18] highlighted the potential severity of this condition, reporting a mortality rate of 12.09%, especially in adults. Conversely, other complications, such as surgical-site infection, wound dehiscence, and seroma, were noted at very low rates. Importantly, no cases of postoperative bleeding or anastomotic leakage were identified in their analysis.[18]

CONCLUSION

Littre's hernia remains an exceedingly rare entity in the pediatric population, with its diagnosis being almost invariably intraoperative due to nonspecific clinical and radiological presentations. The presence of a mixed Littre's hernia mandated segmental intestinal resection with primary anastomosis, in addition to the definitive hernia repair. An associated undescended testis required an orchidopexy as an essential third component of the procedure.

This case report underscores the critical need for a thorough intraoperative assessment, individualized surgical decision-making, and the necessity of considering unusual concurrent urogenital anomalies when managing pediatric inguinal hernias. Favorable long-term outcomes affirm the effectiveness of a comprehensive and tailored surgical approach in this complex patient group.

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Conflict of interest

None

Authorship

- Yudeilys Rodríguez Ávalos: Conceptualization, data curation, research methodology, writing (original draft), review and edition.
- Yesika Muiños Hernández: Supervision, validation, review and edition.
- Enrique Loret de Mola Pino: Visualization, review and editing.

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