



Littre's hernia: a systematic review of the literature

D. Schizas¹ · I. Katsaros¹ · D. Tsapralis² · D. Moris³ · A. Michalinos¹ · D. I. Tsilimigras¹ · M. Frountzas¹ · N. Machairas⁴ · T. Troupis⁵

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Abstract

Purpose A hernia containing Meckel's diverticulum is called a Littre's Hernia. It's a rare entity and its diagnosis is often incidental during routine hernia repair surgery. The objective of this study is the evaluation of the current evidence on Littre's hernias regarding their clinical presentation and optimal treatment approach.

Methods PubMed and Cochrane bibliographical databases were searched from the beginning of time (last search: August 1st, 2018) for studies reporting on Littre's hernias in adult population.

Results Forty-five studies met our inclusion criteria and reported collectively on 53 patients (21 males and 32 females) presenting at health care units with a Littre's hernia. The most common sites of occurrence were femoral (39.6%) and inguinal (34%). The vast majority of cases (77.4%) concerned incarcerated hernias. All patients underwent surgical hernia repair accompanied by a diverticulectomy and 16.9% of them received mesh. Only 7.5% of patients experienced immediate postoperative complications.

Conclusions A Littre hernia is a rare complication of Meckel's diverticulum. It requires surgical attention and all medical professionals should be encouraged to report such cases to expand our experience and optimize the therapeutic approach.

Keywords Littre's hernia · Meckel's diverticulum · Rare hernias · Incisional hernias

Introduction

A Littre hernia is defined by the presence of Meckel's diverticulum in the hernia sac [1]. At the beginning of the 18th century French physician and anatomist, Alexis de Littre, originally reported ileal diverticula and attributed them to traction [2, 3]. In 1785 August Gottlieb Richter proposed

their congenital existence and later, in 1809, Johann Friedrich Meckel studied the embryology of their development [3]. Finally, Sir Frederic Treves described the differences between a Littre and a Richter hernia [4].

Meckel's diverticulum, being present in about 2% of adult population, is one of the commonest congenital anomalies of the gastrointestinal tract [5]. It is usually found on the anti-mesenteric border of the ileum, 20–90 cm from the ileocecal valve [6–8]. It usually presents no specific symptoms and only around 4% of the patients, having a Meckel's diverticulum, experience related complications. These include gastrointestinal bleeding, bowel obstruction, inflammation and perforation [9, 10]. The leading complication in adults is considered to be hemorrhage, due to the presence of heterotopic gastric mucosa [11]. It is followed in frequency by small-bowel obstruction, which may be a result of an external or internal hernia [12].

The existence of Meckel's diverticulum in a hernia sac is quite rare and its exact frequency still remains unknown [13]. A Littre hernia is usually presented as an inguinal, umbilical or femoral hernia [4, 7, 8]. Its symptomatology is similar to any other hernia containing small intestine and

✉ I. Katsaros
gikats13@gmail.com

¹ First Department of Surgery, National and Kapodistrian University of Athens, Laikon General Hospital, Ag. Thoma 17 str., Goudi, 15771 Athens, Greece

² Surgical Department, General Hospital of Ierapetra, Ierapetra, Greece

³ Department of Surgery, Duke University Medical Center, Durham, NC, USA

⁴ Third Department of Surgery, National and Kapodistrian University of Athens, Attikon University Hospital, Athens, Greece

⁵ Department of Anatomy and Surgical Anatomy, National and Kapodistrian University of Athens, Athens, Greece

as a result its diagnosis is regularly made intraoperatively. The ileal loop, to which the Meckel diverticulum is attached, usually follows in the hernia sac and may become incarcerated or even strangulated [14].

The objective of this article was to systematically review the current evidence of published studies reporting on Littre hernias in adult population and evaluate their clinical presentation and treatment approach.

Methods

This systematic review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta Analyses (PRISMA) guidelines [15]. A search of PubMed and Cochrane bibliographical databases was carried out for eligible articles (last search: August 1st, 2018) using all possible combinations of the following keywords: “Littre”, “vitelline duct”, “omphalomesenteric duct”, “meckel diverticulum” and “hernia”. Title and abstract screening was conducted independently by two investigators (D.S., I.K.) using Abstrackr tool [16]. Furthermore, all the references of relevant articles were checked using snowball technique. A third reviewer (D.T.) resolved any occurring disagreements.

In this systematic review we included all English-language articles reporting on Littre hernias in adults. We defined as Littre hernias the ones containing Meckel’s diverticulum in the hernia sac. Articles not written in English, referring to children or autopsy specimens, not presenting a treatment approach and letters to the editor were excluded from this systematic review.

Data were extracted regarding age, sex, and symptoms of the patients. Time interval between symptoms onset and hospital admission was also recorded. Type of Littre hernia (true, mixed), specific location, characteristics and the occurrence of bowel obstruction were also accumulated. We defined as “true Littre hernias” the ones containing only Meckel’s Diverticulum and as “mixed Littre hernias” the ones where Meckel’s Diverticulum is accompanied by small intestine or other abdominal viscera. Additionally, we collected data concerning, type of treatment, mesh use, 30-day posttreatment complications and period of hospitalization.

Furthermore, a statistical analysis of the outcomes was performed by tabulating and then analyzing them using IBM SPSS Statistics for Windows, Version 24.0. Armonk, NY: IBM Corp.

Results

The literature search yielded 345 articles. Forty-five studies met our inclusion criteria and were included in this systematic review. The trial flow diagram is shown in Fig. 1.

Included studies were published from 1954 [17] to 2018 [18] with the majority of them being published after 2008. They reported collectively on 53 patients presenting to healthcare units with 46 true and 7 mixed Littre hernias. Summarized demographic characteristics of all cases included are presented at Table 1.

More specifically, included studies reported on 21 (39.6%) males and 32 (60.4%) females with a mean age of 60.25 ± 16.71 (mean, SD). Their symptoms included abdominal pain, distention, nausea, vomiting. Median time interval between symptoms onset and admission to hospital was 2 days (range 5 h to 11 days). Twenty-one patients (39.6%) suffered from femoral hernias and 18 (34.0%) from inguinal hernias. Other hernia sites include umbilical (6 cases), obturator (3 cases), spigelian (2 cases) and three ventral abdominal hernias. Seven of the cases referred to postoperative hernias. Forty-one hernias (77.4%) were incarcerated with 24 (45.3%) of them being strangulated and 5 (9.4%) perforated. Eighteen patients (34.0%) presented with symptoms of bowel obstruction. Table 2 shows hernia characteristics per site of occurrence.

All patients underwent a hernioplasty combined with a diverticulectomy. Fifty (94.3%) received an open surgical treatment and 3 (6.8%) had a laparoscopic approach. Mesh was used in the repair of 9 hernias. Four patients (7.5%) experienced postoperative complications, including wound dehiscence and minor surgical site infection. One patient died on 3rd day following surgery, due to multiple organ failure following a perforated obturator hernia [19]. Hospitalization period ranged from 1 to 32 days with a median of 6 days. Seventy-five percent of the patients remained in the hospital less than 10 days.

Discussion

A Littre hernia is a rare complication of Meckel’s diverticulum (MD) and it is a result of its protrusion through a herniary orifice. Its incidence is yet unknown, but is reported that 1% of patients having a MD will develop a Littre hernia [18, 20]. It should be distinguished from Richter hernia, where a part of the intestinal wall is strangulated in the hernial sac, but no MD is involved [21]. To our knowledge this is the biggest systematic review concerning Littre hernia in adult population.

A Littre hernia containing only MD is called a true Littre hernia, whereas the simultaneous presence of small intestine or other abdominal viscera in the hernia sac justifies a mixed Littre hernia [22]. Our findings suggest that the overall incidence of true Littre hernias is about seven times greater than the one of mixed Littre hernias. Obturator hernias were the only site that mixed hernias were the majority (66.7%) of the cases with MD being herniated along with small bowel

Fig. 1 Trial flow diagram of this systematic review

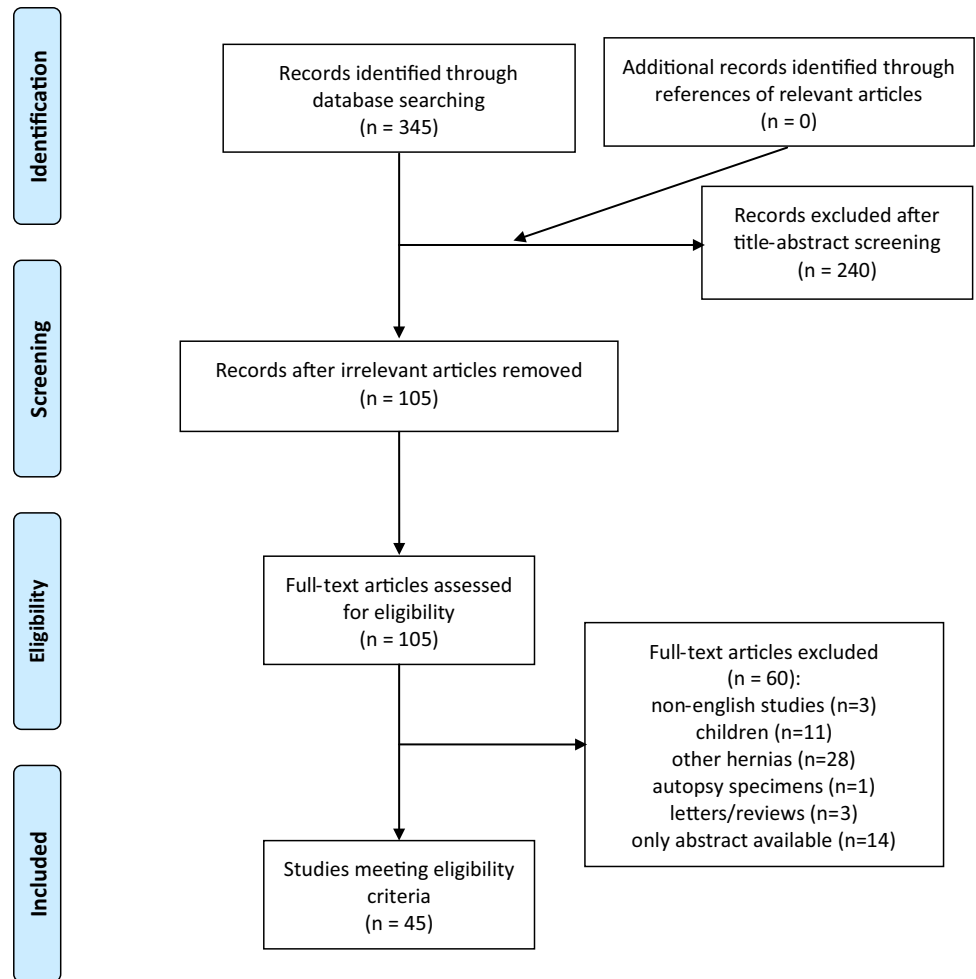


Table 1 Demographic characteristics of included cases

Hernia site	Cases [N (%)]	Age (mean ± SD)	Females [N (%)]	True Littre hernias [N (%)]
Umbilical	6 (11.3%)	52.30 ± 15.20	5 (83.3%)	6 (100%)
Inguinal	18 (34.0%)	54.89 ± 19.35	5 (27.7%)	17 (94.4%)
Femoral	21 (39.6%)	67.24 ± 12.35	16 (76.1%)	17 (80.1%)
Obturator	3 (5.7%)	73.33 ± 2.89	3 (100.0%)	1 (33.3%)
Spigelian	2 (3.8%)	59.5 ± 23.34	1 (50.0%)	2 (100.0%)
Ventral abdominal hernia	3 (5.7%)	46.67 ± 14.30	2 (66.7%)	3 (100.0%)
Total	53 (100.0%)	60.25 ± 16.71	32 (60.4%)	46 (86.7%)

SD standard deviation

loops [19, 23]. Another interesting finding was the concurrent presence of MD and appendix in two femoral hernia cases [20, 24].

Meckel’s diverticulum is a congenital intestinal blind pouch consisting of all intestinal layers and is present in 0.3–3% of adult population [5, 10, 25]. It is the embryologic remnant of the omphalomesenteric duct and arises from its incomplete obliteration during the 5th week of gestation [22,

26, 27]. Its clinical image ranges from completely asymptomatic to complicated causing gastrointestinal bleeding, bowel obstruction, inflammation, and perforation [9, 10, 12]. Age younger than 50 years, male sex, diverticulum length greater than 2 cm and the presence of histologically abnormal tissue predispose to symptomatic MD in adults [11]. A well differentiated neuroendocrine tumor and a microscopic carcinoid were found during the histological examination

Table 2 Hernia characteristics per occurrence site

Hernia site	Cases (N)	Incarcerated [N (%)]	Strangulated [N (%)]	Perforated [N (%)]	BOWEL obstruction [N (%)]
Umbilical	6	4 (66.7%)	2 (33.3%)	1 (16.7%)	2 (33.3%)
Inguinal	18	10 (55.5%)	7 (38.9%)	1 (5.6%)	4 (22.2%)
Femoral	21	20 (95.2%)	12 (57.1%)	1 (4.8%)	6 (28.6%)
Obturator	3	3 (100.0%)	2 (66.7%)	2 (66.7%)	3 (100%)
Spigelian	2	2 (100.0%)	0 (0.0%)	0 (0.0%)	1 (50%)
Ventral abdominal hernia	3	2 (66.7%)	1 (33.3%)	0 (0.0%)	2 (66.7%)
Total	53	41 (77.4%)	24 (45.3%)	5 (9.4%)	18 (34.0%)

of surgically resected MD present in Littre hernias [28, 29]. Heterotopic gastric tissue was present at two of our included cases and one additional case pertained unspecified ectopic tissue [30–32]. All symptomatic Meckel’s diverticula should be resected, but there is still controversy regarding the resection of incidentally found asymptomatic MD [1, 8, 14, 33].

The diagnosis of a herniated Meckel’s diverticulum is usually made intraoperatively [34]. The role of both abdominal ultrasonography and computed tomography (CT) is crucial, but frequently they do not reach a definite diagnosis [19, 35, 36]. Additionally, plain radiographs can reveal signs of intestinal obstruction, but rarely unveil its cause [19]. In our systematic review, ultrasonography was utilized in eight cases and CT in ten cases to support diagnostic procedure. Nonetheless, the presence of MD in the hernia sac was confirmed during surgery in all cases.

Littre hernias are usually presented as inguinal, femoral and umbilical hernias [4, 37]. Our findings are in accordance with that, but the incidence of each location is different to the ones reported by Skandalakis et al. [4]. In our included studies femoral and inguinal hernias stood for 39.6% and 34% of the cases, respectively. Moreover, umbilical hernias were present at 11.3% of the cases and obturator hernias at 5.7%. We also found two cases (3.8%) of spigelian hernias and three cases (5.7%) of ventral abdominal hernias.

Although Meckel’s diverticulum is more frequently encountered in men, Littre hernias occur more often in women, mainly due to the high incidence of femoral and obturator Littre hernias [11, 25, 38–40]. Our findings concluded that 60.4% of the cases concerned females. Male patients stood for the 72.3% of the inguinal hernias and 50% of the spigelian hernias, while women were the majority concerning all the remaining hernia sites.

A Littre hernia demonstrates atypical signs and symptoms including abdominal pain, distention, nausea, and vomiting. Its progress is more gradual compared to other hernias [22, 41]. In our study patients admitted to health care units 5 h to 11 days (median 2 days) after the onset of their symptoms. Common complications include incarceration,

strangulation, and perforation [7, 42]. Racy et al. report that 1 out of 680 strangulated femoral hernias and 4 out of 654 strangulated inguinal hernias contain a Meckel’s diverticulum [20]. Sometimes, even in cases of incarcerated Littre hernias, there is no intestinal obstruction present, as only MD is “trapped” and the rest bowel is free, which is a similarity with Richter’s hernia usual clinical presentation [21, 30]. Perforation may be the result of either peptic ulceration related to gastric acid or compromised circulation related to strangulation [7, 22, 38]. In our study, almost 8 out of 10 of hernias were incarcerated, 45.3% of them strangulated and 9.4% perforated. Eighteen incarcerated hernias resulted in bowel obstruction. The majority of femoral and obturator hernias were strangulated. On the contrary no spigelian hernia presented signs of strangulation. Only one (5.6%) inguinal hernia was perforated.

The repair of a Littre hernia consists of both hernia repair and removal of Meckel’s diverticulum [8]. All patients received an open (94.3%) or laparoscopic (5.7%) hernia approach followed by a MD resection. Mesh was applied only in 17% of the cases, while the remaining ones had a suture repair. The presence of incarceration or perforation and the possible field contamination often make difficult the use of mesh [43, 44]. The fact that over 30% of the patients were treated before 1994 should also be considered for the interpretation of the results, as surgeons were not accustomed to the use of mesh during routine hernia repairs. Over 90% of the patients experienced no postoperative complications and the majority of them were dismissed from the hospital in less than 10 days. The remaining patients experienced postoperative complications, such as wound dehiscence and minor surgical site infection. One elderly woman died on 3rd postoperative day, due to a perforated obturator hernia and respiratory failure, that resulted in multiple organ failure and sepsis [19].

In conclusion, Littre hernia is any hernia containing Meckel’s diverticulum. Despite being a rare entity, it can be a possible finding during a routine investigation of any hernia and warrants surgical attention. A careful examination

of any hernia sac should be performed by the operating surgeon. Furthermore, all surgeons should not only be aware of this rare type of hernia, but are also encouraged to consistently report such cases to enhance available literature regarding best clinical management of Littre's hernia.

Author contributions Conception and design: DS, NM. Data collection, analysis and interpretation: DS, IK, DT, TT. Writing the manuscript: DS, IK, DT, MF, AM. Critical revision of the manuscript: DS, IK, DM, DIT.

Compliance with ethical standards

Conflict of interest DS declares no conflict of interest. IK declares no conflict of interest. DT declares no conflict of interest. DM declares no conflict of interest. AM declares no conflict of interest. DIT declares no conflict of interest. MF declares no conflict of interest. NM declares no conflict of interest. TT declares no conflict of interest.

Ethical approval Approval from the institutional review board was not required for this type of study.

Human and animal rights This article is a systematic review and it contains data collected through literature review. It does not include research directly involving human or animal participation.

Informed consent For this systematic review, informed consent was not necessary.

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