

Littré's Femoral Hernia Causing Intestinal Fistula

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ABSTRACT: Littré's hernia is one of the rarest forms of hernia, characterized to contain the Meckel's diverticulum as its sole content. The Meckel's diverticulum in the hernial sac can be involved by those complications which occur in the diverticulum in the free abdominal cavity.

A case of Littré's femoral hernia in a 75 years old man with formation of an intestinal fistula as a result of inflammation of the Meckel's diverticulum, probably, following mechanical trauma exerted by a phytogenic fibre, was reported, and the previous literatures were reviewed. Consequently, it was confirmed that signs and symptoms of Littré's hernia with complications are less severe and non-characteristic compared to ordinary types, and hence its preoperative diagnosis is very difficult.

KEY WORDS: Meckel's diverticulum, intestinal fistula, femoral hernia, phytogenic fibre, diverticulectomy, Littré's hernia.

INTRODUCTION

The eponymous nomenclature, such as Littré's or Richter's hernia, is sometimes confusing. Alexis Littré described two cases of hernia with an intestinal diverticulum in 1700, and, later, cases of hernia with intestinal wall as well in 1714. Independently, in 1785, A. G. Richter reported a lesion in which only a part of the intestinal wall was strangulated. Consequently, the terms, Littré's and Richter's hernia, have often been used synonymously. In order to avoid confusion, it seems to be appropriate to adopt the definition suggested by Pabst¹⁶ or Keynes²⁶, *i.e.* hernia with the Meckel's diverticulum as its sole content is defined as Littré's hernia, while those with the intestinal wall Richter's hernia.

Although the Meckel's diverticulum is not a rare anomaly, Littré's hernia as defined is fairly rare. Hence, a case of Littré's femoral hernia, with an intestinal fistula formation, is reported in this communication and the current literatures are reviewed.

Report of a case: A 75 year old man with an intestinal fistula of about ten months standing was first seen at the outpatient clinic of the Hitachi General Hospital, Ibaragi, on 12th October, 1970. The patient noticed a slightly tender mass of about the size of sparrow's egg in his right femoral region for the first time around September, 1969, when the mass was easily reduced by gentle massage without any bowel sound. On 31st December, 1969, after having overeaten, he felt epigastric fullness and lost appetite. On 2nd January, 1970, the mass slightly enlarged and became harder and was found to be irreducible. Since then, his abdomen gradually distended. On 9th January

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the mass became tender, and on the following day the overlying skin became hyperemic and edematous with local fever.

He visited a local doctor on 10th January, who, detecting neither signs of intestinal obstruction nor those of peritoneal irritation, made a diagnosis of simple acute subcutaneous abscess probably caused by lymphadenitis and decided to evacuate it. A 5 cm long incision was laid across the centre of the mass in parallel to the inguinal ligament. Necrotic granulation tissue without pus was exposed and biopsied, which was reported to be non-specific acute lymphadenitis later. The wound was packed with gauze intending for secondary healing. Contrary to expectation, the wound would not close, and the discharge increased in amount turning out to be dirty yellow like intestinal content. Local X-ray examination revealed a fistulous tract leading to the terminal ileum. After continuous discharge for about four months, the fistula closed spontaneously, but recurred after about one month with signs of acute inflammation around the previous fistulous orifice. Since then, the spontaneous closure and reopening of the fistula alternated in every two or three weeks.

Since September, 1970, the patient became aware of abdominal distention with borborygmus, and sometimes with vigorous bowel movements. Throughout the course, he neither nauseated nor vomited. On admission, he was found thin and slightly anemic, but his general condition did not deteriorate despite of a long-standing intestinal fistula. His abdomen was distended with visible peristalsis in the right and lower abdomen, sometimes with audible borborygmus. There was a diffuse swelling around the previous incisional scar. The overlying skin was slightly hyperemic and edematous, and a little hyperpigmented. The central part of the scar, about 5 mm in diameter, was retracted showing the previous orifice of the fistula. Diffuse subcutaneous induration was palpated corresponding to the visible swelling which extended up to the right inguinal region. The skin over the swelling was neither tender nor warm compared with the neighboring area. No definite femoral canal was palpable. The fistulous tract was obliterated, and there were no signs to suggest the existence of intestine closely beneath the skin. Besides hypertrophic prostate, the pelvic floor was slightly indurated and tender. Both inguinal canals were normal. All the laboratory findings were within the normal limit.

Radiological examinations: A plain roentgenogram of the abdomen in the upright position showed a loop of the small intestine distended with air in the right abdomen without fluid level. Barium meal reached this distended loop four hours after ingestion, where it was mixed with air, and vigorous backward flashing with audible bowel sound was observed. Two hours later, the contrast medium passed the Bauchin's valve located about 80 cm distal to the loop. The ileum about 50 cm oral to the Bauchin's valve was tightly adherent to a site near the previous incisional scar, and the intestine kinked so sharply at the site to form a check valve that the proximal segment of the intestine was distended. The terminal ileum was slender without any pathological finding. A short slender tail-like projection was seen hanging down from the bending point (Fig. 1). A tentative diagnosis of Richter's femoral hernia causing intestinal fistula was made basing on the clinical findings.

On 11th November, 1970, a hernioplasty was performed under general anesthesia to restore patency of the intestine. The lesion was proved to be typical femoral hernia, and was treated after the modified Zimmermann's method by dividing the inguinal ligament. The hernial sac was found to be firmly adherent to the previous scar, and a

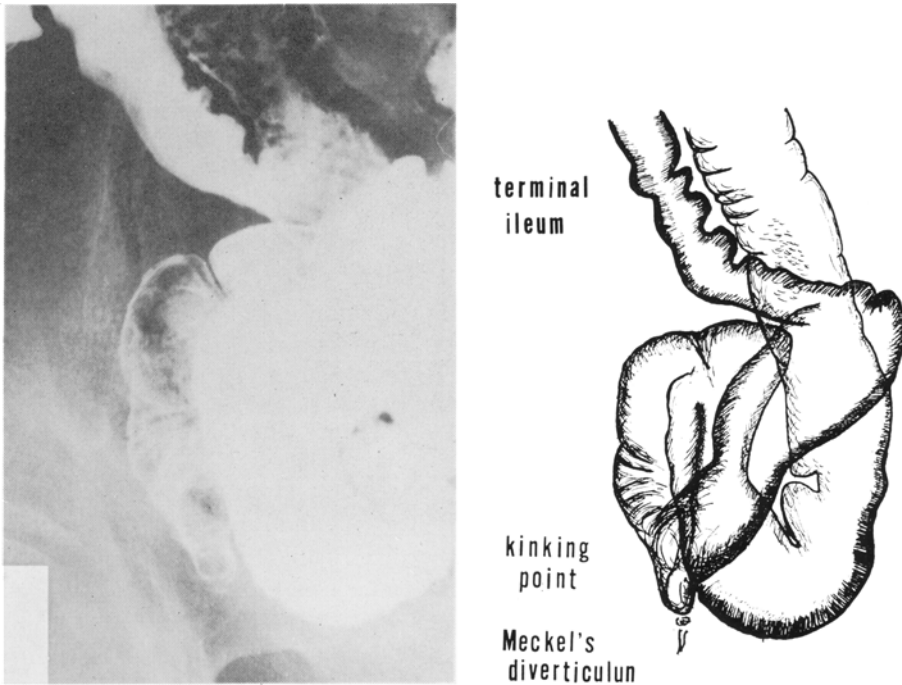


Fig. 1.—Schematic drawing of Littré's hernia causing intestinal obstruction by kinking.

conglomeration of the femoral lymph nodes was seen fixed to the medial aspect of the sac, which was felt as a movable firm mass. The Meckel's diverticulum, about 3 cm in length and 1 cm in diameter, was found to be the sole content of the hernial sac. The serosa of the diverticulum was fibrotically thickened. The Plica serosa and the neighboring serosa of the diverticulum were tightly adherent to the ventrolateral aspect of the sac. The tip of the diverticulum submerged into the fibrous scar to be united to the scar and the previous fistulous tract. The femoral canal was about 2.5 cm in diameter,



Fig. 2.—Cut surface of the resected ileum with Meckel's diverticulum on the antimesenteric aspect.

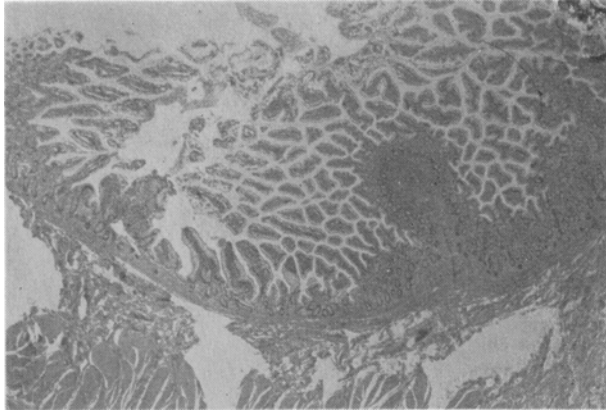


Fig. 3.—Section of the Meckel's diverticulum stained with hematoxylin and eosin. $\times 60$

through which the adjoining ileum could be pulled out, and this part of the intestine had the fibrotically thickened serosa as well. The diverticulum, the scar tissue containing the previous fistulous tract with femoral lymph nodes, and about 8 cm of the ileum in continuity with the diverticulum were resected *en bloc*. The stumps of the ileum were approximated with an end-to-end anastomosis. The patient's postoperative course was in general uneventful.

Pathology of the resected specimen: A conical Meckel's diverticulum, 3×1 cm, was seen on the antimesenteric aspect of the resected ileum. A piece of skin and conglomerated lymph nodes surrounded by fibrous scar tissue were seen at the tip of the diverticulum (Fig. 2). The muscle layer of the diverticulum was thickened and contracted. Macroscopical examination of the specimen did not disclose any ulceration or erosion of the mucous membrane, tumor, foreign body and other lesions in the lumen of the diverticulum. Histological sections of the diverticulum showed a thin mucous membrane with short villi. The number of crypt cells was scantier than the corresponding part of the ileum, but goblet cells were abundant, and a few Paneth cells were seen at the bottom of the crypt (Fig. 3). The tip of the diverticulum fused into the subcutaneous

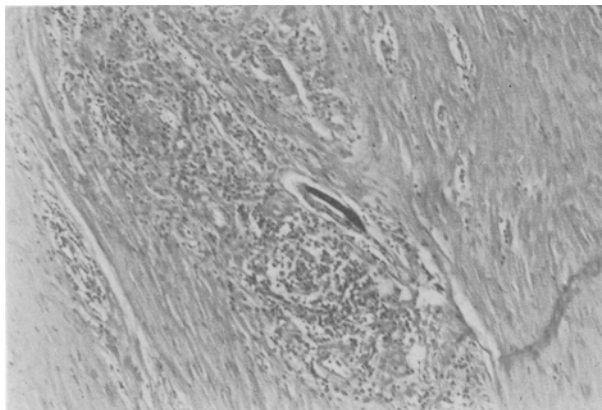


Fig. 4.—Foreign body in granulation tissue at the tip of the diverticulum. $\times 150$

fibrous granulation tissue, and a piece of foreign body, probably tough phytogenic fibre, was seen in the fibrous granulation tissue (Fig. 4). The cellular components of the inflammatory infiltration in the granulation tissue were mainly composed of lymphocytes with scattered eosinophiles. Fibroblasts and histiocytes were also abundant. Neither heterotopic gastric nor pancreatic tissue was seen in the diverticulum. Ulceration or erosion in the mucous membrane of the diverticulum was not demonstrated either. The lymph nodes attached to the apex of the diverticulum showed signs of non-specific chronic inflammation.

DISCUSSION

The incidence of the Meckel's diverticulum in general population has been reported to be between 0.5 to 2 per cent in the West^{1,4,11,16,20} as well as in Japan^{49,65,29}. Previous publications have revealed that about 15 to 20 per cent of all Meckel's diverticula bear some sorts of complication^{4,9,19}. Aside from diverticulitis, hemorrhage from the ulcerative lesion due to the heterotopic gastric mucosa is frequent in the West^{1,9,14,19,23}, in contrast to Japan, where frequent occurrence of intestinal obstruction caused by this structure has been reported rather than hemorrhage^{34,43,46,71,72}.

Table 1.—Association of Meckel's diverticulum with different hernias in Japan compared with the report of Pabst

	Reports in Japan		Report of Pabst	
Inguinal	20:	16M, 4F. Rt.: 11M, 1F. Lt.: 4M, 2F. Uk.: 1M, 1F.	68:	47M, 11F, 10Uk. Rt.: 32M, 8F, 6Uk. Lt.: 10M, 3F, 1Uk. Uk.: 5M, —, 3Uk.
Femoral	9:	1M, 6F, 2Uk. Rt.: 1M*, 5F, 1Uk. Lt.: —, 1F, —. Uk.: —, —, 1Uk.	22:	6M, 15F, 1Uk. Rt.: 4M, 7F, 1Uk. Lt.: 1M, 4F, —. Uk.: 1M, 4F, —.
Umbilical	2:	1M, —, 1Uk.	25:	5M, 5F, 15Uk.
Cicatrical	2:	1M, —, 1Uk.	—	— — — —
Internal	—	— — — —	1:	1M, —, —.
Unknown	2:	—, 1F, 1Uk.	7:	1M, —, 6Uk.
Sum	35:	19M, 11F, 5Uk.	123:	60M, 31F, 32Uk.

Rt. = Right, Lt. = Left, Uk. = Unknown, M. = Male, F. = Female.

* = present case.

Footnotes to the Table 1

References for the cases reported in Japan;

Inguinal,

27, 30, 31, 33, 36, 40, 41, 44, 47, 50, 51, 53, 54, 58(2), 59, 62, 66, 69, 74.

Femoral,

32, 35, 37, 48, 55, 56, 63, 73.

Umbilical,

52, 70.

Cicatrical,

64, 67.

Unknown,

28, 57.

It is fairly rare to find the Meckel's diverticulum in hernia³, but the diverticulum can fairly often be seen in hernia complicated with intestinal fistula^{7,15,18,25}. Among 1,040 cases of hernia (922 of inguinal, 21 of femoral and 97 of other sorts of hernia) operated on at the First Department of Surgery, Tokyo University Hospital and Hitachi General Hospital from April, 1963, to December, 1970, the present case was the only one belonging to Littré's hernia.

Littré's hernia has been reported to occur more often in inguinal hernia than in other sorts of hernia.¹⁸ The cases collected from the Japanese literatures have also shown the same trend (Table 1). In order to explain the reason for this tendency, by citing two cases which showed fibrous adhesion between the capsule of the testis or epididymis and the diverticulum, Ekehorn formed a theory that the Meckel's diverticulum adhered to the retroperitoneum was pulled into the inguinal canal during the course of *Descensus testis*⁶. However, as it is not rare to find fibrous adhesions between the hernial sac and the greater omentum or other abdominal viscera, temporary circulatory disturbance or irritation to the structure in the sac causes fibrin deposit on the serosal surface resulting in the formation of collagenous fibrous adhesions between them. As the Meckel's diverticulum can be seen in the femoral or abdominal cicatricial hernia, which is secondary in origin, the existence of the diverticulum in the hernial sac is not necessarily congenital, but is the phenomenon brought forth quite by chance. Anyhow, it is true that the Meckel's diverticula are more often seen in the state adherent to the hernial sac than in the free state¹⁶.

Non-specific diverticulitis^{9,19}, peptic ulceration of the mucous membrane caused by heterotopic gastric glands,^{1,14,22,42} fat necrosis caused by acute inflammation of the aberrant pancreas^{12,61}, necrosis of tumor⁴, strangulation of the diverticulum⁸, mechanical trauma caused by enterolith^{2,20}, fish bone^{46,60}, seed or parasite^{39,68} have been reported to cause destruction of the diverticular wall. When these changes do occur in the diverticulum in hernial sac, they can result in fistula formation after involving the whole layer of the diverticulum extending into the sac and further to the overlying skin. The possible cause of intestinal fistula formation in the present case can be due to diverticulitis induced by mechanical trauma owing to intruded phytogenic fibres into the diverticulum. As there was a considerable slackness around the diverticulum in the femoral canal, inflammation seems to be mainly responsible for the fistula formation. However, the possibility of other intestinal segment having taken part together with the Meckel's diverticulum in causing elastic incarceration of the diverticulum at the onset can not be denied.

Mildness of symptoms, in spite of severe necrotic inflammation occurred in hernia leading to perforation of the diverticulum should be emphasized in the present case. In accordance with the first description of Littré cited in the previous literatures^{6,16,25,26}, the following points seem to be pathognomonic:

- 1) No change in bowel movement.
- 2) Seldom occurrence of hiccup.
- 3) No or seldom vomiting in contrast to ordinary incarcerated hernia.
- 4) No distension of the abdomen.
- 5) Later appearance of tumor in the groin, which tends to be small. Inflammatory signs and other symptoms appearing later and in milder way.

As incarcerated Littré's hernia causes inflammatory changes without severe abdomi-

nal symptoms, the lesion can likely be misdiagnosed as acute lymphadenitis. It would be more so in case of examining the demented senile for the first time who is not likely to offer any helpful informations.

Richter's hernia is nearly always found in the incarcerated state, but Littré's hernia is sometimes confirmed quite by chance. The Meckel's diverticulum in hernia is said to be more often inflamed than strangulated²⁵. Since the intestine does not usually show any signs of obstruction providing more than one third of the lumen is intact^{17,26}, intrusion of the diverticulum into the hernial sac alone does not necessarily result in intestinal obstruction. In the present case, the diverticulum became fixed to the fundus of the sac to cause kinking of the intestine from which it arose, thus inducing intestinal obstruction (Fig. 1).

The terminal ileum is the site where duplication of the intestine of various kinds is often found. Those lesions belonging to the split notochord syndrome are usually found in the mesenteric aspect of the intestine³⁸, but the other sorts of duplication are difficult to be differentiated from each other. The structure lacking the *Plica serosa* or remnants of the *Vasa omphalomesenterica*, especially the one arising not from the antimesenteric aspect of the intestine is difficult to be defined as the Meckel's diverticulum.

Relatively high incidence of complications in the Meckel's diverticulum has raised a trend to remove the structure whenever possible^{1,9,19}. In order to avoid possible occurrence of subsequent ileo-ileal intussusception after diverticulectomy, Aubrey advocated a method to resect the diverticulum by clamping its neck transversely against the intestinal axis and suture the cut edges in two layers¹. Resection of the diverticulum with adjacent ileum followed by end-to-end anastomosis can be done without any difficulties in the adult.

Incarceration of the greater omentum, herniation of the appendix or appendicitis in hernial sac, subcutaneous abscess, acute lymphadenitis, inflamed *Hydrocele funiculi spermatici*, etc. are to be differentiated from complicated Littré's hernia. Are there really specific features for complicated Littré's hernia facilitating to differentiate the lesion from the others? According to Pabst, Nauman¹⁶ described the characteristics of Littré's hernia as the following^{16'}:

- 1) Pain occurs in a small circumscribed area.
- 2) The abdomen may be distended with tenderness around the navel, but, the ileocaecal area is free from tenderness.
- 3) Pain may be localized around the navel.
- 4) Vomit may be bile-stained soon after the onset of the accident, but it seldom becomes feculent.
- 5) Obstipation with abdominal distress may have been repeated, such distress may recur after long intervals. The patient may have experienced colicky pain with vomiting.
- 6) The abdomen may distend in spherical shape.
- 7) Meteorism may be prominent.
- 8) Malformation may coexist in other parts of the body.

Referring these characteristics, Pabst reviewed 123 cases of Littré's hernia. As he could not reconfirm the above mentioned features in those cases, he concluded that it was impossible to differentiate Littré's hernia from the others basing on symptoms observed in each of the cases^{16'}. Thus correct preoperative diagnosis of *Hernia Littrica*

is extremely difficult. However, one must bear this entity always in mind during surgical exploration, and the Meckel's diverticulum must be removed, whenever possible, to avoid potential serious complications in future.

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