Digital Edition e-publish on-line ISSN 2239-253X

Direttore Nicola Picardi

# Mesothelial cyst of the round ligament of the uterus



A case report of a rare condition

## Beatrice D'Orazio\*/\*\*, Giovanni Corbo\*/\*\*, Guido Martorana\*\*\*, Gaetano Di Vita\*, Girolamo Geraci\*

\*General Surgery Unit, Department of Surgical, Oncological and Stomatological Sciences, University of Palermo, Italy \*\*Postgraduate Medical School in General Surgery, University of Palermo, Palermo, Italy \*\*\*General and Oncological Surgery Unit, Fondazione Istituto G. Giglio, Cefalù, Italy

### Mesothelial cyst of the round ligament of the uterus. A case report of a rare condition

BACKGROUND: Differential diagnosis of inguinal mass must include, especially in female patients, a wide variety of lesions among which our analysis will focus on mesothelial cyst of the round ligament of the uterus. A rare developmental lesion often misdiagnosed as hernias and accidentally detected during surgical exploration of the groin region.

CASE REPORT: Of a left inguinal mass causing local discomfort and progressive worsening of local pain. A pre-operative diagnosis of left symptomatic femoral hernia was made and the patient consented to surgical treatment. The surgical exploration of the inguinal and femoral canals revealed a femoral hernia associated to a clear fluid cystic lesion of around 2 cm arising from the round ligament. Histopathology demonstrated a mesothelial cyst of the round ligament

CONCLUSIONS: Mesothelial cysts of the round ligament of the uterus must be taken into consideration in the differential diagnosis of groin swelling in female patients and a greater effort is needed in order to reach a preoperative diagnosis and prevent an over treatment.

Key words: Mesothelial cyst, Preoperative diagnosis, Uterus

#### Introduction

Mesothelial cyst of the round ligament of the uterus is a rare developmental cause of groin mass in women, not often considered in the differential diagnosis of groin swelling and usually identified at the time of a surgical exploration  $^{1}$ .

Differential diagnosis of inguinal mass must include:

- inguinal or femoral hernia;
- lymph nodes;
- parasitic infections;
- vessels abnormalities;
- endometriosis;
- round ligament cystic lesions;
- benign or malignant soft tissue tumors.

Especially in female patients it should include endometriosis and round ligament cystic lesions <sup>2</sup>.

#### Case Report

A 57 years old, multiparous, smoker female with a BMI of 37 kg/m<sup>2</sup>, presented at our department for a 2 months history of a left inguinal mass causing discomfort, heaviness sensation and progressive worsening of local pain. The clinical examination revealed a mass at the left femoral region, of around 3cm, which could be palpated during coughing and Valsalva's manoeuvre. There weren't any associated systemic symptoms, nor local signs of inflammation, such as erythema or oedema, or intestinal obstruction signs. Laboratory tests were normal. Hence, a pre-operative diagnosis of left symptomatic femoral hernia was made and the patient consented to surgical treatment.

We performed the procedure under local anaesthesia. The surgical exploration of the inguinal and femoral canals revealed a femoral hernia associated to a cystic lesion of

*Ann Ital Chir, 2020; 9 -* Sept. 28 pii: S2239253X2003337X Online Epub

Pervenuto in Redazione Maggio 2020. Accettato per la pubblicazione Giugno 2020

Correspondence to: Gaetano Di Vita, General Surgery Unit, Department of Surgical, Oncological and Stomatological Sciences, University of Palermo. Via Liborio Giuffrè 5 - 90127 Palermo, Italy (e-mail: divitagaetano@libero.it)



Fig. 1: Intraoperative photograph of the mesothelial cyst originating from the left round ligament.

around 2 cm protruding in close proximity to the external inguinal ring (Fig. 1). Hence, we proceeded with the alloplastic repair of the hernia, by a plug tension free technique, and the excision of the cystic lesion.

The post operatory recovery was uneventful and the patient was discharged the same day.

Histopathology (Fig. 2) demonstrated a mesothelial cyst of the round ligament of  $2.5 \times 2$  cm filled with seromucinous clear ambered fluid, lined with single layer of cuboidal cells; immunochemistry was positive to calretinin confirming the mesothelial origin.

After 6 months of ultrasonographic follow up we didn't observe signs of recurrence at the left groin nor any abnormalities at the right side.

#### Discussion

Round ligament of the uterus is a derivative of gubernaculum and attaches paramesonephric duct at 9 week's <sup>3</sup>, it extends caudally through the inguinal canal to the labioscrotal swelling forming a fibrous band during the 3<sup>rd</sup> trimester; the Nuck's canal is a portion of peritoneum which carries some layers of the abdominal wall to be incorporated in the round ligament <sup>4</sup>. Hence the round ligament of the uterus originates at both uterine horns, it leaves the pelvis through the deep inguinal ring and inguinal canal to reach the labium majum <sup>5</sup>. A mesothelial cyst of the round ligament is a very rare entity and can occur at any point of its path <sup>5</sup>. This is a developmental disorder for the origin of which 3 main hypothesis have been formulated 6; the first one is based on the flowed obliteration of Nuck's canal 7, the second on the inclusion of embryonic, mesenchymal and mesothelial elements or remaining during the development of the ligament <sup>2</sup>, the third and last one, that is only a



Fig. 2: Calretin stain (IHCx100): section of the mesothelial cyst filled with seromucinous clear ambered fluid, lined with single layer of cuboidal cells; immunochemistry was positive to calretinin

speculation for now, relies on the genuine development of the cyst as benign cystic mesothelioma <sup>8</sup> which could be reactive to pelvic inflammation or endometriosis or neoplastic with a real malignant potential <sup>9</sup>.

Only 10 cases have been described in the literature from 1980 to 2013 <sup>6</sup>, the first knowledge of this entity dates 1854 <sup>2</sup> but only Ponka <sup>10</sup> in 1980 described it precisely as a mesothelial cyst of the round ligament for the first time.

Therefore, this rare cystic lesion is often misdiagnosed, thus its actual incidence may be much greater than in literature  $^4$ .

Generically this lesion occurs in women in their  $3^{rd} - 4^{th}$  decade <sup>3</sup>, in 82% of the cases it appears at the right side <sup>5</sup>, from 30% up to 50% of cases it is found to be associated to inguinal hernia <sup>11</sup>. The pre-operative diagnosis is a real challenge as for its graded and unspecific clinical features; these lesions are often asymptomatic or characterized by subtler symptoms compared to hernias such as pain, local discomfort or heaviness sensation <sup>3-7</sup>. The imaging study of choice for the differential diagnosis of these cystic lesions is Ultrasound of the inguinal region, this become mandatory every time we face an unexpansible groin mass. In our case, the high BMI allowed us to appreciate the femoral hernia but not the associated cystic lesion.

Usually the definitive diagnosis is macroscopic and histological <sup>6</sup>, as a matter of fact they are usually accidentally detected during groin exploration of the inguinal region for herniorrhaphy <sup>2</sup>. From the histological point of view they lined with a single layer of flat, cuboidal cells with the characteristics of mesothelial cells <sup>4</sup>.

Due to their benign nature, the surgical excision is indicated only if the cysts are symptomatic or quickly increasing in volume, otherwise an Ultrasound follow up seems to be adequate  $^{3}$ .

#### Conclusions

In light of the above, it is clear that a mesothelial cyst of the round ligament of the uterus should be taken into consideration in the differential diagnosis of an asymptomatic groin swelling in female patients <sup>12</sup>. Therefore, we conclude by asking if an US (ultrasound) study of the inguinal region in those patients should be considered necessary in order to reach a proper pre-operative diagnosis and avoid an over treatment of this cystic lesions.

#### Riassunto

Le cisti mesoteliali del legamento rotondo sono una rara causa di tumefazione inguinale nelle donne, la loro eziopatogenesi rimare poco chiara e i casi riportati in letteratura sono scarsi. Inoltre, quest'ultime sono raramente prese in considerazione nella diagnosi differenziale delle tumefazioni della regione inguinale nelle pazienti di sesso femminile, ragion per cui sono spesso identificate solo in corso di esplorazione chirurgica e correttamente diagnosticate solo dopo esame istopatologico.

Una donna di 57 anni (BMI 37 kg / m<sup>2</sup>) si è presentato nel nostro dipartimento con una massa inguinale sinistra, con storia di 2 mesi, che causava disagio locale e peggioramento progressivo del dolore locale. È stata fatta una diagnosi preoperatoria dell'ernia femorale sintomatica sinistra e la paziente ha acconsentito al trattamento chirurgico. L'esplorazione chirurgica dei canali inguinale i e femorale ha rivelato un'ernia femorale associata a una chiara lesione cistica fluida di circa 2 cm derivante dal legamento rotondo. L'istopatologia ha dimostrato una cisti mesoteliale del legamento rotondo CONCLUSIONI: Le cisti mesoteliali del legamento rotondo dell'utero devono essere prese in considerazione nella diagnosi differenziale del gonfiore inguinale nelle pazienti di sesso femminile e sono necessari maggiori sforzi per raggiungere una diagnosi preoperatoria e prevenire un trattamento eccessivo.

L'indicazione chirurgica per queste lesioni rimane legata alla presenza di sintomi invalidanti, data la natura benigna delle stesse, per cui, al fine di evitare un overtreatment, ci sembra utile sottolineare innanzitutto, come questa entità patologica debba essere presa in considerazione nel percorso diagnostico di una tumefazione inguinale in soggetti di sesso femminile e in secondo luogo, l'utilità dell'esame ecografico di routine in queste pazienti.

#### References

1. Habibi M, Kazak M, Arioz Habibi H, Bulbuller N: *A rare cause of inguinal mass: round ligament cyst.* Pol Przegl Chir, 2018; 90: 47-52.

2. Harper GB Jr, Awbrey BJ, Thomas CG Jr, Askin FB: *Mesothelial cysts of the round ligament simulating inguinal hernia. Report of four cases and a review of the literature.* Am J Surg, 1986; 151:515-57.

3. Ryley DA, Moorman DW, Hecht JL, Alper MM:A mesothelial cyst of the round ligament presenting as an inguinal hernia after gonadotropin stimulation for in vitro fertilization. Fertil Steril, 2004; 82:944-46.

4. Oh SN, Jung SE, Lee JM, Chung JH, Park GS: Sonographic diagnosis of a round ligament cyst in the inguinal area. 2007; 35:226-28.

5. Saylam B, Gülseren MO, Han O, Comçali B, Vural V, Coşkun F: *Cysts of the round ligament simulating inguinal hernia: Report of a case.* J Nippon Med Sch, 2013; 80:296-99.

6. Manatakis DK, Stamos N, Agalianos C, Vamvakas P, Kordelas A, Davides D: *Mesothelial cyst of the round ligament misdiagnosed as irreducible inguinal hernia.* Case Rep Surg, 2013; 2013:408078. doi: 10.1155/2013/408078.

7. Stickel WH, Manner M: Female hydrocele (cyst of the canal of Nuck): sonographic appearance of a rare and little-known disorder. J Ultrasound Med, 2004; 23:429-32.

8. Uzüm N, Ozçay N, Ataoğlu O: Benign multicystic peritoneal mesothelioma. 2009; 20:138-41.

9. Tentes AA, Zorbas G, Pallas N, Fiska A: *Multicystic peritoneal mesotheli*oma. Am J Case Rep, 2012; 13:262-64.

10 Ponka JL: *Hernias of the Abdominal wall.* Philadelphia: WB Sounders 1980; 119-20.

11. Kim BM, Lee JY, Han YH, Kim SY, Seo JW, Kim YH, Cha SJ, Hur G, Joo M, Lee ES: *Mesothelial cyst of the round ligament mimicking a metastasis: A case report*. Korean J Radiol, 2010;11:364-67.

12. Apostolakis S, Ioannidis A, Koutserimpas C, Patrikakos P, Perrakis A, Velimezis G: *Mesothelial cyst of the round ligament*. G Chir, 2018; 34: 323-25.