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Giant Peritoneal Inclusion Cyst of the Abdominopelvic Cavity, a Very Rare Case Report and Literature Review

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Abstract The objective of this article is to present a rare case such as peritoneal inclusion cyst or a benign cyst mesothelioma, a rare injury that predominantly affects women of reproductive age, with a still unknown etiology. Materials and Methods: We present the clinical case of a 36-year-old women, asymptomatic, with a pelvic ultrasound reported free abdominal fluid in a medical checkup. In her medical history revealed that six years ago the patient underwent laparotomy for complicated appendicitis with peritonitis. The abdominal and pelvic scan showed a giant cystic lesion of 20 cm x 17 cm x 10 cm, which occupies part of the abdomen and pelvis. Result: surgical treatment was decided and the patient underwent laparotomy, achieving the complete elimination of cystic mass and small bowel resection joined the cyst was performed. The histopathology report confirmed the diagnosis of peritoneal inclusion cyst. Conclusion: location, type of injury and behavior of mesothelioma cysts should be considered as they have a high recurrence rate and are often asymptomatic.

Keywords: *peritoneal inclusion cyst, benign cystic mesothelioma, laparotomy*

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1. Introduction

Mesentery cysts are rare lesions with an incidence of 1 in 100,000 adult patients. In 1970, Mennemeyer and Smith by using electron microscopy defined the differences between multicystic mesothelioma and abdominal lymphangioma.

In 2000, Perrot et al proposed a new classification of mesenteric cysts based on immunohistochemistry reports that are now currently the most widely accepted: [1]

2. Mesenteric Cysts Classification

1. **Linfatic origin cyst**
   A. Simple linfatic cyst
   B. Lymphangioma
2. **Mesothelial origin cyst**
   A. Simple cystic mesothelial
   B. Benign cystic mesothelioma
   C. Malign cystic mesothelioma
3. **Enteric cyst origin**
   A. Duplication intestinal cyst
   B. Enteric cyst
4. **Urogenital cyst**
5. **Dermoid cyst**
6. **Nonpancreatic pseudocyst**
   A. Traumatic origin
   B. Infectious origin. [1]

Peritoneal inclusion cysts (PIC) are a very rare lesion that affects the peritoneum with a predominant affection for the pelvis; since 2004, there have been 130 cases reported worldwide, and in 2015, we found a total of 173 cases worldwide; due to this, this is an extremely rare pathology in the world literature Usually it affects a perimenopausal women, between 30 to 40 years old; although there's usually a history of pelvic surgery, pelvic inflammatory disease or endometriosis [2]; the etiology currently remains uncertain. Is often asymptomatic and its imaging characteristics represent a dilemma for both radiologists and surgeons. PIC management varies from observation with serial imaging, hormones, image guided aspiration with or without sclera therapy and surgical excision to complete resection.

3. Clinical Case

We present a multiparous 36 year old female patient, with a surgical history of a laparotomy 6 years ago due to appendicitis with secondary peritonitis; patient was referred by her gynecologist because a routine checkup found free abdominal liquid in an abdominal ultrasound. Patient is referred to an oncological surgeon for a surgery. It should be emphasized that the patient was asymptomatic.

Physical exam reveals a soft, painless and mobile mass located in the lower abdominal cavity; CBC and blood

chemistry are within normal ranges, where there's a slight elevation of CA 125 marker of 395 u / ml. (<35 u / ml).

The CT report shows a defined, hypodense image, with liquid density in the retroperitoneum and pelvic cavity, displacing the bowel loops in which the contrast medium won't enhance, the dimension of the mass is 20 x 17 x 10 cm; there's a thickening of the visceral mesenteric fat which also contains free liquid that's observed in the pelvic cavity. (Figure 1)

A laparotomy was performed, which we found an unilocular cystic mass that measured 25 cm x 20 cm x 18 cm, which extends from the pelvis to the lower abdomen, with bowel and colon displacement; it also extends to the inframesocolic retroperitoneum without a well-defined but transparent and thin wall. In the interior of the cyst a black-green liquid (1800 ml), was also found without partitions inside (Figure 2).

In addition, areas of fibrosis that appear to be cancerous in the small intestine was observed, at 60 cm and 120 cm from the ileocecal valve, infiltrating the intestinal wall measuring 3 cm x 2 cm and 5 x 2 cm. (Figure 3).

Extensive adhesiolysis and cystic tumor excéresis was performed, including resection of 80 cm of the terminal ileum and peritonectomy with a complete omentectomy, and end to end anastomosis of the terminal ileum.

The content of the giant cystic mass was drained and sent to intraoperative cytology, which reported a benign liquid

Pathology Report: a cyst wall composed of lax connective tissue, that's partially encased by a reactive mesothelium type and patches of mixed inflammatory infiltrate; the serous layer of the small intestine showed a marked fibrosis and giant-cellular reaction due cholesterol crystals along with foreign bodies and surgical suture thread; the final diagnosis was a Peritoneal Inclusion Cyst (Figure 4).

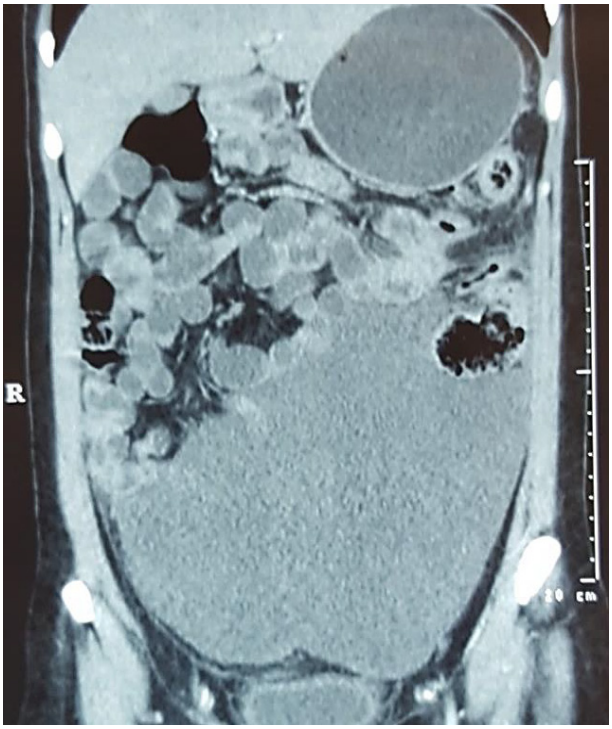

Figure 1. CT scan showing giant cyst (20cm x 17cm x 10 cm)

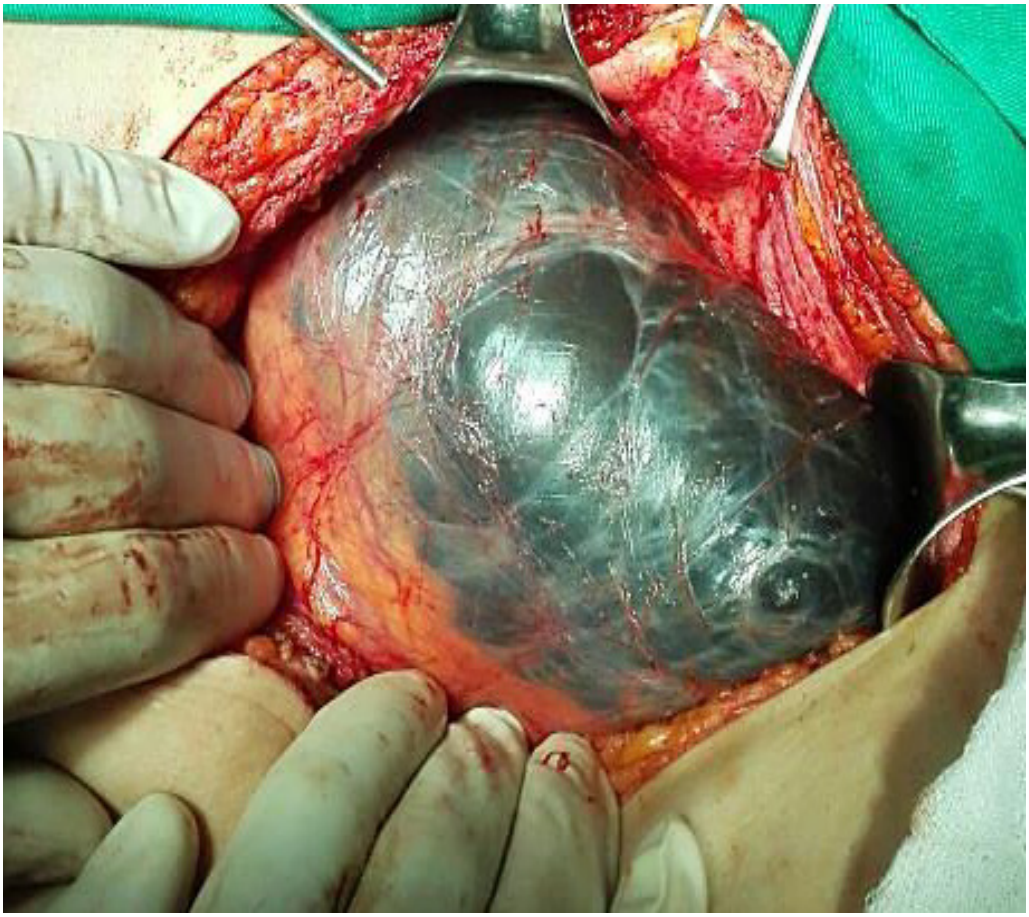

Figure 2. Laparotomy: cyst mass of 25 x 20 cm, with dark green liquid content

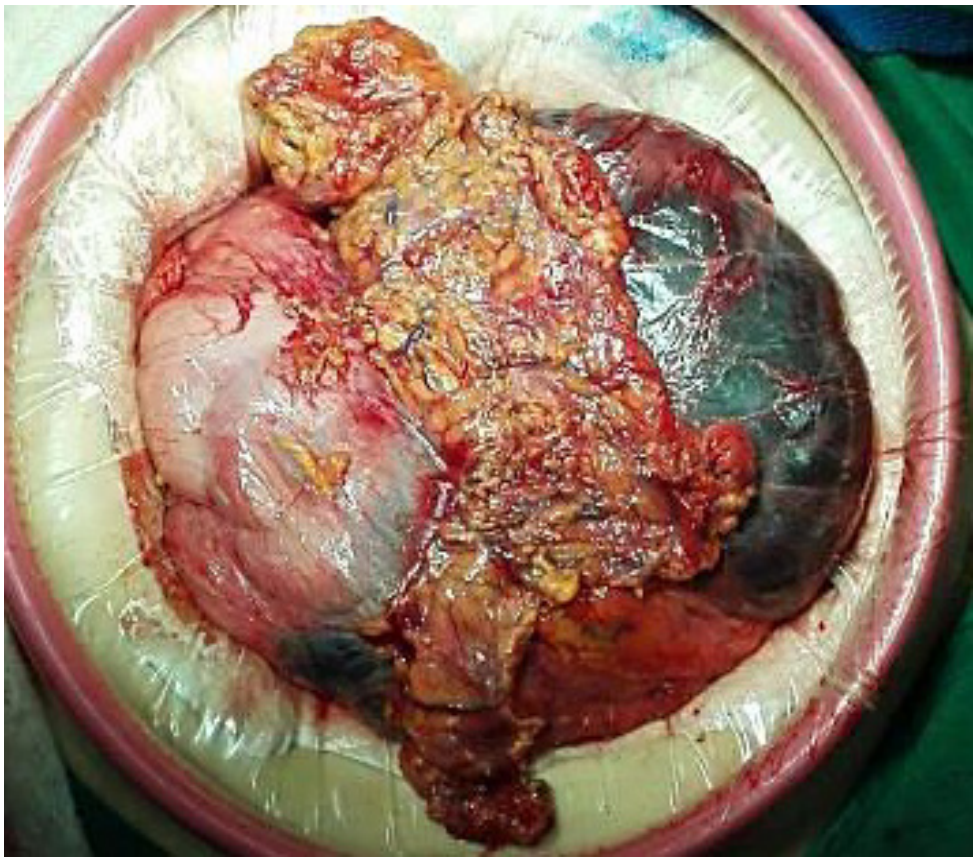

Figure 3. cystic mass adhered to the omentum and small intestine

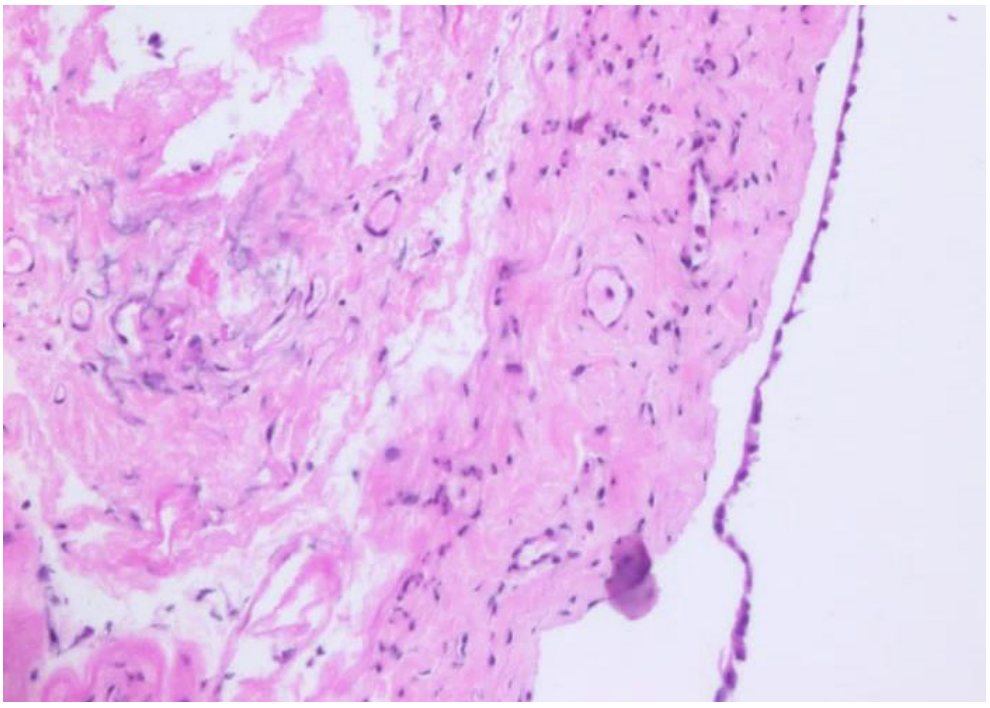

Figure 4. Histopathology: mesothelial cyst wall coating (Metropolitan Hospital Courtesy, Pathology Service)

No postoperative complications, with satisfactory evolution.

4. Discussion

The peritoneal mesothelium gives rise to rare lesions, whether they're benign, malignant or borderline in origin, these can be confused by the physician performing the initial diagnostic approach.

Peritoneal tumors are classified as smooth muscle, epithelial and mesothelial tumors, as well as peritoneal tumors of unknown origin. Mesothelial tumors are classified as benign cystic mesothelioma, adenomas, as well as differentiated papillary mesothelioma and malignant peritoneal mesothelioma. [3]

One of the rarest peritoneal tumors known is the benign cystic mesothelioma, also known as "peritoneal inclusion cyst", "cystic peritoneal mesothelioma" and "post-operative peritoneal cyst" due to its prior history with surgical procedures.

In 1889, Henke described this anomaly for the first time and called it a "multiple cystic lymphangioma", in 1979 Mennemeyer and Smith determined the mesothelial origin; and Moore et al. in 1980, dubbed it as a benign cystic mesothelioma. [4]

In 2001, Colina et al. reported 130 cases in women and 19 cases in men. [3] In 2006, Safioleas's found only 130 cases described. In 2015, Cosme da Silva et al. found a total of 173 cases worldwide. [5]

The peritoneal inclusion cyst is a tumor found between the benign adenoma tumor, and undetermined peritoneal mesothelioma. [6]

4.1. Presentation Form

These lesions can present in different forms: a) as a solitary cystic mass (as in our case), b) as a solitary multicystic mass, c) as multiple cystic lesions scattered throughout the peritoneum.

It occurs most frequently in premenopausal women and is associated with a history of pelvic inflammatory disease, abdominal surgery, or endometriosis. However, there are a few cases reported in male patients, where the risk of recurrence and malignancy is likely higher. [7]

Although the process mostly affects the peritoneum, they have also been reports of affections in other cavities, such as the pleura, pericardium and spermatic cord. [6]

Although it is a benign disease, it causes discomfort and anxiety until a definitive diagnosis is made, due to its size and characteristics. Telli et al. argue that these lesions have a neoplastic etiology. [8]

It affects women in about 90% of the time, most often between 30 and 40 years of age. Elbouhaddouti et al. reported that the recurrence rate is higher in women: 40 to 50%, whereas in men it is 33%, and with an excellent prognosis; there has been only one documented death reported. Rafat et al. reported a case of a 14 year old patient, who had a partial resection of the abdominal mass, and died 12 years after refusing a new surgery. [9]

Probable causes are: fibers or foreign bodies, inflammatory mediators and mechanical injury. Although a reactive neoplasic cause has also been proposed, genetic or familial inheritance has not been established, except in a few isolated cases associated with familial Mediterranean fever. Mazziotti et al. reported that there may be an association with autoimmune diseases that cause a reaction in the peritoneum involving autoantibodies and inflammatory reactions, such as in Crohn disease. [10]

4.2. Clinical Presentation

Signs and symptoms are nonspecific; abdominal pain common, palpable mass and/or symptoms due to compression of the mass, although some patients remain asymptomatic. The diagnostic approach is done with ultrasound and computed tomography, in which unioculate or multioculate cystic tumors are observed, which are not dependent of the abdominal organs. [11]

4.3. Differential Diagnosis

Although there is countless diagnosis, it is more likely to be confused with multilocular cystic lymphangioma, but this usually occurs in males (75%) and mostly in children under 5 years old (60%). Microscopically, lymphangiomas present as lymphoid aggregates in the smooth muscle of the cystic walls. Problem cases can be differentiated by immunohistochemistry. [12]

4.4. Treatment

The first treatment option is a complete surgical resection. Sometimes, as in this case, when the cyst is compromising and infiltrating the adjacent organs such as the intestine, it is necessary to perform a bloc resection of the tumor along with the cystic mass and the organs involved.

In case of a tumor recurrence, a guided percutaneous drainage by tomography can be done; Birch et al. reported that about 50% of cysts recur after aspiration. In case of a suspected malignancy an open biopsy can be performed. The advantage of surgery is that it provides a definitive diagnosis. A surgical approach may be done by laparoscopy or laparotomy. [13]

4.5. Recurrence

Recurrence rate is 50%, Roen et al. reported a recurrence rate of 27-75%, so strict control is required. [14]

In case of first line treatment or recurrence, it has been proposed that the combination of surgery with intraperitoneal hyperthermal chemotherapy in select patients, a low recurrence rate (16.7%) compared to surgery alone (41.7-50%) has been demonstrated. The prognosis is generally good; with a mean survival of 5 years is 100% with combined treatment. [15]

5. Conclusion

In conclusion, we presented a 36 year old woman with a history of secondary peritonitis that was diagnosed 6 years ago, the patient remains asymptomatic; with an incidental finding of a giant cystic mass; although this is a rare condition, we emphasize the importance of suspected peritoneal inclusion cyst in female patients who are premenopausal with a history of previous surgery. Knowing the clinical associations is useful for diagnosing and treatment, this may permit conservative management in selected cases.

Statement of Competing Interests

The authors declare no conflict of interest.

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