

Retroperitoneal approach for recurrent benign multicystic peritoneal mesothelioma

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Abstract

Benign multicystic peritoneal mesothelioma (BMPM) is an uncommon lesion. It presents as a lump in abdomen or a finding seen on imaging modalities. Surgery is the primary modality of treatment. However, it has a high recurrence rate; this results in adhesions and subsequent surgeries difficult. We present a case of recurrent BMPM in a female operated twice earlier in a rural centre. Imaging modalities showed majority of the lesion in paracolic and retroperitoneal region. Hence, retroperitoneal approach for surgery was taken. This avoided previous surgical adhesions. A brief case report on this novel approach and review of literature is presented.

Introduction

Benign multicystic peritoneal mesothelioma (BMPM) is a rare peritoneal lesion. It commonly presents as lower abdominal pain, palpable mass, or both. It is also known as multilocular peritoneal inclusion cyst. Benign cystic mesothelioma should be included in the differential diagnosis when investigating pelvic masses or abscesses associated with either appendicitis or pelvic inflammatory disease or ovarian cyst in women. These cysts are often found on imaging or incidentally at surgery. These lesions arise from peritoneal mesothelium that covers the serous cavity. They are typically benign with a high recurrence rate as well as the potential for malignant transformation. It is advised to confirm the diagnosis by radiology, histology and immunohistochemistry. We present a case of recurrent multilocular peritoneal inclusion cyst in a 32-year-old female operated twice earlier. The mass was attached to retroperitoneal surface of ascending colon, hence a novel retroperitoneal approach is being described with review of the literature.

Case Report

A 32-year-old female presented with recurrent lump in right lumbar region. There were no associated urinary or bowel complaints. She was operated twice for similar lumps in a rural hospital 10 and 3 years back. Her discharge card of previous surgery showed the diagnosis of lymphangioma. Her case papers of first surgery done 10 years back were not available. On physical examination a lump in right lumbar region just lateral to previous paramedian incision was palpated. Rest of the abdomen was unremarkable.

Ultrasonography showed multicystic complex mass in right lumbar region measuring 25×8×7 cm. Computed tomography scan abdomen and pelvis (Figure 1) showed a 25×10×6.5 cm sized well defined non enhancing lobulated multiloculated cystic lesion in right paracolic gutter on lateral aspect of ascending colon. Inferiorly it was abutting the lateral wall of urinary bladder. Similar morphology lesions were seen in pouch of Douglas and mesentery. All these finding were suggestive of recurrent lymphangioma.

Patient was subjected to surgery with a retroperitoneal approach as majority of the mass was retrocolic in position. Incision was taken below and parallel to 12th rib extending from posterior axillary line and going anteriorly towards umbilicus by keeping the patient in left lateral position. Initially retroperitoneal component of lesion was dissected out from kidney and ureter and lastly from retro peritoneal surface of colon. During this peritoneum was opened and the intra peritoneal component including pelvic component was completely removed. Intra operative finding showed a cystic multiloculated mass loosely attached to serosal surfaces of the ascending colon extending into pelvis with intra peritoneal extension into pouch of Douglas. The rest of the appendix, caecum, large and small bowel appeared grossly normal. Both ovaries and the uterus were visualized and found to be normal. The lesion was friable, hence care was taken to avoid rupture contamination of the pelvis with the cyst fluid. Complete resection of the lesion was performed. The diagnosis was not clear at this point, but our differential included a mucinous cystadenoma or pseudomyxoma peritonei or lymphangioma. As the diagnosis of BMPM was not established hence intraoperative hyperthermic peritoneal infusion with cisplatin was not considered. The specimen was a multicystic mass that was 22 cm wide in its greatest dimension (Figure 2). The individual cysts, which ranged between 3 and 9 cm in diameter, contained serous fluid.

Post operative recovery was uneventful. Histopathology showed cystic spaces lined by flattened cuboidal to hobnail epithelium sepa-

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rated by thin fibrous septa (Figure 3). The lining epithelium showed strong immunopositivity for calretinin (Figure 4) and immunonegativity for CD31. Hence diagnosis of multilocular peritoneal inclusion cyst was confirmed. Follow up of 1 year has shown patient to be disease and symptom free.

Discussion

Peritoneal inclusion cyst was first described in 1979 by Mennemeyer and Smith.¹ It usually occurs in young to middle-aged women with a history of pelvic surgery,² endometriosis^{3,4} or pelvic inflammatory disease.^{5,6} Approximately 150 cases of benign multicystic peritoneal mesothelioma, with various presentations have been reported since 1979.⁷ Peritoneal inclusion cysts represent a non-neoplastic reactive mesothelial proliferation. The lesions range in size from several mm in diameter to bulky masses. The development of a peritoneal inclusion cyst depends on the presence of an

active inflammation and peritoneal adhesions. The normal peritoneum absorbs fluid easily, however when the peritoneum becomes infected or mechanically injured, its transport properties are changed and fluid absorption is slower. This causes a decrease in the clearance of peritoneal fluid. These conditions are believed to interfere with peritoneal re-absorption. This would tend to support a hypothesis of being reactive and inflammatory rather than neoplastic.

Peritoneal inclusion cysts usually occur in women of reproductive age group. Mesothelial cysts may be arranged in grape-like clusters or present as isolated lesions studding the surface of the peritoneum. They often contain more mucinous or gelatinous fluid. Microscopically, they are lined by a single layer of cuboidal or flattened mesothelium that may focally form papillary projections, nests or tubules. Occasionally they undergo squamous metaplasia. There may be mild to moderate cytologic atypia, but no mitoses. The formation of glands that have an infiltrative pattern may be mistaken for cancer. Vacuolated mesothelial cells may easily resemble signet-ring carcinoma. Unlike other cysts of the mesentery and retroperitoneum, cystic mesothelioma has a propensity to recur in up to 50% of cases.

Pathological differential diagnosis includes benign and malignant lesions that present as cystic or multicystic abdominal masses. Benign lesions include cystic lymphangioma (cystic hygroma),^{8,9} which is the most common differential diagnosis, where cells are immunoreactive to vascular markers (CD31, CD34, factor VIII and VEGFR3). Another important one is pseudomyxoma peritonei which is the result of mucinous adenocarcinoma of appendix, presenting as a mucocele. Rare forms include cystic forms of endosalpingiosis,^{10,11} endometriosis,¹² mullerian cysts involving the retroperitoneum,¹³ cystic adenomatoid tumors¹⁴ and cystic mesonephric duct remnants.¹⁵ Malignant lesions include malignant mesothelioma and serous tumors involving the peritoneum.⁶

In our case, immunohistochemistry showed strong positivity for calretinin and negative for CD31, which is a vascular marker. Thus diagnosis of multilocular peritoneal inclusion cyst was confirmed.

The high chances of recurrences even if resection margins are clear is due to widespread peritoneal seedlings. Currently palliative surgical debulking with reoperations for recurrence is the mainstay of treatment. However, some have advocated sclerosive therapy with tetracycline, continuous hyperthermic peritoneal perfusion with cisplatin, and antioestrogenic drugs. The optimal treatment may be cytoreductive surgery with peritonectomy combined with perioperative intraperitoneal chemotherapy with cisplatin to elimi-

nate all gross and microscopic disease.¹⁶ The goal of this treatment regimen is to reduce the likelihood of progression or recurrence. In present case, the diagnosis was confirmed only after histology and immunohistochemistry of the specimen, hence intraoperative therapy was not given.

Open midline approach has been described in literature, however in a recurrent disease with predominant retroperitoneal component; a retroperitoneal approach can be useful. In the present case, this novel approach helped in dissecting the lesion from kidney, ureter and duodenum. Moreover, it could be easily sepa-

rated from the underlying surface of ascending colon. The intraperitoneal component could also be assessed and removed by opening the peritoneum laterally. This resulted in complete excision with minimal handling of the bowel.

Conclusions

Common problem associated with BMPM is high chance of recurrences even if resection margins are clear, due to widespread peritoneal seedlings. Currently palliative surgical debulking with reoperations for recurrence is the mainstay of treatment. In our case, the mass being attached to retroperitoneal surface of ascending colon, hence a novel retroperitoneal approach was used. This resulted in minimal bowel handling and avoided peritoneal seedlings. Moreover, the intraperitoneal component could also be removed by opening the peritoneum. This helped in complete excision of the lesion. Hence, this approach may be useful in recurrent BMPM, where majority of mass is retrocolic in position.

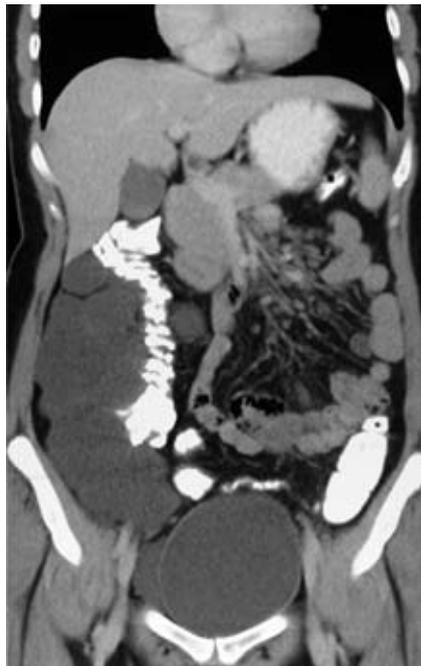


Figure 1. Computed tomography scan abdomen and pelvis showing recurrent benign multicystic peritoneal mesothelioma as paracolic mass.

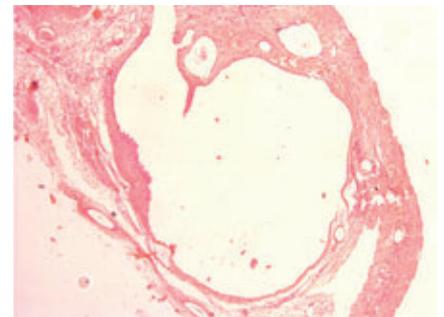


Figure 3. Microscopic view showing multicystic nature of the tumor lined by mesothelial cells (hematoxylin and eosin stain 200×).

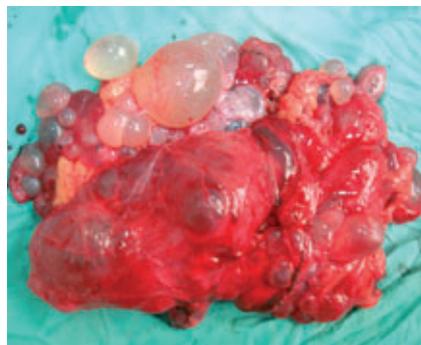


Figure 2. The specimen of benign multicystic peritoneal mesothelioma.

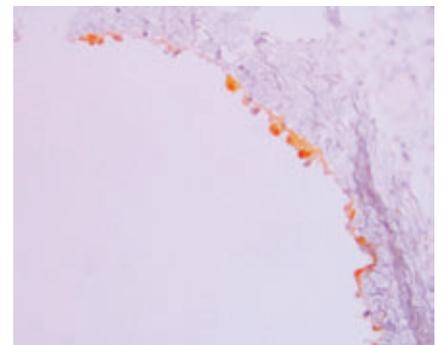


Figure 4. Immunohistochemistry showing Calretinin positive (400×).

References

1. Mennemeyer R, Smith M. Multicystic, peritoneal mesothelioma: a report with electronmicroscopy of a casemimicking intra-abdominal cystic hygroma (lymphangioma). *Cancer* 1979;44:692-8.
2. McFadden DE, Clement PB. Peritoneal inclusion cysts with mural mesothelial proliferation. A clinicopathological analysis of six cases. *Am J Surg Pathol* 1986;10:844-54.
3. Groisman GM, Kerner H. Multicystic-mesothelioma with endometriosis. *Acta Obstet Gynecol Scand* 1992;71:642-4.
4. Urbanczyk K, Skotniczny K, Kucinski J, Friediger J. Mesothelial inclusion cysts (so-called benign cystic mesothelioma) - a clinicopathological analysis of six cases. *Pol J Pathol* 2005;56:81-7.
5. Tangjitgamol S, Erlichman J, Northrup H, et al. Benign multicystic peritoneal mesothelioma: cases reports in the family with diverticulosis and literature review. *Int J Gynecol Cancer* 2005;15:1101-7.
6. Ross MJ, Welch WR, Scully RE. Multilocular peritoneal inclusion cysts (so-called cystic mesotheliomas). *Cancer* 1989;64:1336-46.
7. Snyder JA, Carman Jr R, Aggon AA, Cardinale JP. Benign multicystic peritoneal mesothelioma: A rare case presenting as pneumoperitoneum and pneumatosis intestinalis. *J Gastrointest Oncol* 2011; 2:55-8.
8. Carpenter HA, Lancaster JR, Lee RA. Multilocular cysts of the peritoneum. *Mayo Clin Proc* 1982;57:634-8.
9. Katsube Y, Mukai K, Silverberg SG. Cystic mesothelioma of the peritoneum: a report of five cases and review of the literature. *Cancer* 1982;50:1615-22.
10. Clement PB, Young RH. Florid cystic endosalpingiosis with tumor-like manifestations: a report of four cases including the first reported cases of transmural endosalpingiosis of the uterus. *Am J Surg Pathol* 1999;23:166-75.
11. Heatley MK, Russell P. Florid cystic endosalpingiosis of the uterus. *J Clin Pathol* 2001;54:399-400.
12. Bell DA, Scully RE. Benign and borderline serous lesions of the peritoneum in women. *Pathol Annu* 1989;2:1-21.
13. De Peralta MN, Delahoussaye PM, Tornos CS, Silva EG. Benign retroperitoneal cysts of mullerian type: a clinicopathologic study of three cases and review of the literature. *Int J Gynecol Pathol* 1994;13:273-8.
14. Bisset DL, Morris JA, Fox H. Giant cystic adenomatoid tumour (mesothelioma) of the uterus. *Histopathology* 1988;12:555-8.
15. Weiss SW, Tavassoli FA. Multicystic mesothelioma. An analysis of pathologic findings and biologic behavior in 37 cases. *Am J Surg Pathol* 1988;12:737-46.
16. Sethna K, Mohamed F, Marchettini P, et al. Peritoneal cystic mesothelioma: a case series. *Tumori* 2003;89:31-5.