Case Report: Benign Multicystic Peritoneal Mesothelioma

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Benign multicystic peritoneal mesothelioma (BMPM) is a rare tumor of unknown pathogenesis that occurs mainly in women in their reproductive ages and mostly diagnosed intraoperatively. In this paper, a case was summarized with pelvic mass which was handled as an ovarian cancer preoperatively.

A 40 year old woman admitted to gynecologic oncology unit due to pelvic pain. On bimanual gynecologic examination a unilateral, semi-fixated pelvic mass in the right adnexial region was palpated. She had no medical history of malignancy. On pelvic ultrasound in the right adnexial region a multiseptated mass was reported. Ca-125 level was 178.2 IU/ml. On gross examination during operation, there was a thin walled multicystic pelvic mass adherent to posterior cul de sac, rectal serosa, and right pelvic wall. Pathologists' first impression was that mass was not containing any malignant component on frozen sections. Carefull resection of pelvic mass, total abdominal hysterectomy, right salphingooophorectomy and appendectomy performed. The final pathologic diagnosis was BMPM with parafine blocks.

In conclusion BPMP is a rare benign cystic tumor which can be easily misdiagnosed as an ovarian cancer preoperatively. Intraoperative findings and appearence of the mass may mimic malignancy. For that reason frozen section examination will prevent overtreatment.

Key Words: Benign multicystic peritoneal mesothelioma, Peritoneal cyst

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Introduction

Benign multicystic peritoneal mesothelioma (BMPM) is a rare tumor of unknown pathogenesis that occurs mainly in women in their reproductive ages and mostly diagnosed intraoperatively.[1-3] In this paper we summarized a case of BMPM which's preoperative and intraoperative impression was malignant.

Case Report

A 40 year old woman, gravida 2 para 2, admitted to gynecologic oncology unit due to pelvic pain. On physical examination a unilateral, semi-fixated pelvic mass in the right adnexial region which can not be discriminated from uterus clearly was palpated. In her history it was noted that she had pelvic pain for ten days and she had mild vaginal discharge. She admitted to another center and treated for pelvic infection ten days ago. She had no medical history of malignancy. She had an abdominal surgery for cesarean section 17 years ago. On pelvic ultrasound in the right adnexial region and Douglas pouch a multiseptated mass which was adherent to intestinal loops was reported. Ultrasonographic findings resembled an ovarian malignancy (Figure I).

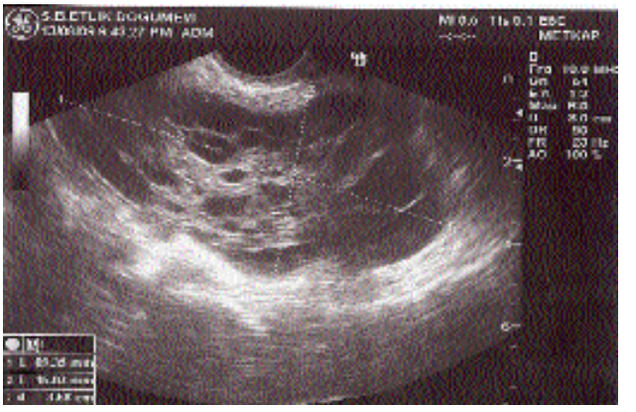

Figure 1: Ultrasonographic images of adnexial mass of the patient.

Ca-125 level was 178.2 IU/ml. Complete blood counts, other tumor markers, and laboratory tests were in normal ranges. The patient underwent a laparotomy. On gross examination there was a thin walled multicystic pelvic mass adherent to posterior cul de sac, rectal serosa, and right pelvic wall. Grape like cysts were 0.5-1cm in size. Frozen sections of this

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mass was reported as small, thin walled, translucent cysts containing serous fluid within (Figure 2). The mass was adherent to the uterus. Pathologists' first impression was that mass was not containing any malignant component and they advised examination of parafine blocks. Carefull resection of pelvic mass, total abdominal hysterectomy, right salphingooophorectomy and appendectomy performed. After surgery patient was discharged from hospital uneventfully at postoperative day 2. Patient was seen at a postoperative first week visit again she has no complaint.

Figure 2: Small, thin walled, translucent cysts containing serous fluid. (X4)

The pathologic diagnosis was BMPM with dimensions of 5x5x3cm. The lesion was not originating from uterus or ovary. Parafine blocks examination was reporting cysts were lined with cuboidal mesothelial cells those were flattened at some points (Figure 3). Cystic structures were separated by fibrous septas. There were bleeding points and inflamatory cells between these septas. Mesothelial cells were containing dark nuclei but there were no atypia. These mesothelial cells were stained positively with vimentin.

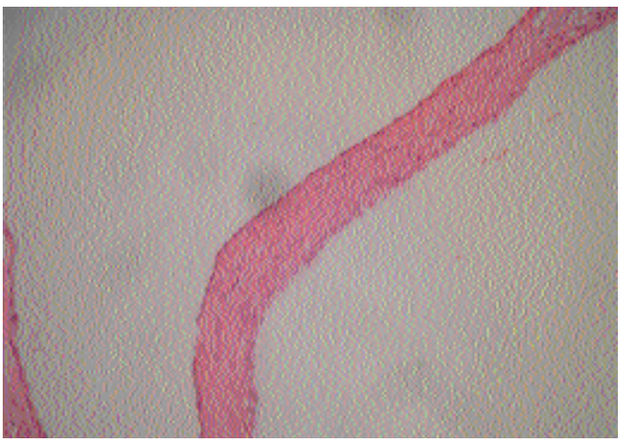

Figure 3: Flattened cuboidal mesothelial cells lining cyst wall. (X40)

Discussion

BMPM is a rare tumor of unknown pathogenesis that occurs mainly in women in their reproductive ages.[1-3] The tumor, which occurs most frequently in young to middle-aged women, affects chiefly the pelvic peritoneum particularly the cul de sac, uterus, and rectum.[1] Clinically the most frequent complaints are chronic pain, a palpable mass and distension of the abdomen, but BMPM can also present as an acute abdomen or may be even asymptomatic.[1-3] Some authors have regarded BMPM a neoplasm, while others argue that it is a reactive proliferation of mesothelium as a result of previous abdominal surgery, pelvic inflammatory disease or endometriosis.[1,5] Association with previous surgery, endometriosis, pelvic inflammatory disease, diverticulosis, and Familial Mediterranean Fever suggests the lesion is a reactive process.

As in this case BMPM is an entity that is difficult to diagnose preoperatively. Although imaging techniques such as ultrasonography, computerized tomography and magnetic resonance imaging can demonstrate the lesion in most cases differential diagnosis from ovarian malignancies or other adnexial pathologies is not possible preoperatively. Preoperative fine needle aspiration biopsy of cystic lesion may help diagnosis.[5,6] The diagnosis can be confirmed by electron microscopy and immunohistochemistry.

History of the patient may include some clues for the differential diagnosis. As in this case most of the patients are positive for an abdominal surgery history.[7] Ravidranauth et al. reported 7 cases with a history of previous abdominal surgery or other preceding intra-abdominal events in 17 patients (41%).

Treatment of BMPM is surgery. Complete resection of the cystic lesion is the only option to prevent recurrence. Although it has a high recurrence rate even after surgical treatment, it does not present a tendency to transform into malignancy. As in this patient many BMPM cases misdiagnosed as other pelvic pathologies such as pelvic infections, benign adnexial pathologies even as ovarian malignancies. In emergency settings surgical procedures performed for BMPM treatment may be very agressive. Also there are experimental treatment options for BMPM but medical treatments with antioestrogens gonodotropin releasing hormone analogues and intraperitoneal chemotherapy have a higher recurrence rate.[8,9]

Since BPMP is most commonly seen in woman in their reproductive ages the extend of surgery is important. Cytoreductive surgery with peritonectomy is the option but hysterectomy and bilateral salpingooforectomy is really necessary? There is not a consensus about this in the literature. Fertility sparing will be the goal during surgery of this benign condition.

In conclusion BPMP is a rare benign cystic tumor which

can be easily misdiagnosed as an ovarian cancer preoperatively. Although it has a benign character it may recur even after complete resection. In these patients intraoperative findings may give an impression of the mass is malignant. At this point frozen section examination will prevent overtreatment. New studies for understanding the pathogenesis of the disease may help prevention the disease and they may also help to determine the definitive treatment options.

Olgu Sunumu: Benign Multikistik Peritoneal Mezetelyoma

Benign multikistik peritoneal mezetelyoma (BMPM) başlıca reprodüktif çağda görülen, çoğunlukla intraoperatif olarak tanımlanan, patogenezi bilinmeyen nadir bir tümördür. Bu çalışmada preoperatif dönemde over kanseri tanısıyla müdahale edilmesi planlanan pelvik kitlesi olan olgu özetlendi.

Kırk yaşında olan hasta pelvik ağrı nedeniyle jinekolojik onkoloji ünitesine kabul edildi. Bimanuel jinekolojik muayenede sağ adneksiyel alanda ünilateral ve semi-fikse pelvik kitle palpe edildi. Hastanın özgeçmişinde malignite öyküsü yoktu. Pelvik ultrasonografide sağ adneksiyel alanda multiseptalı kitlenin varlığı teyit edildi. Ca125 seviyesi 178,2 IU/ml'ydi. İntraoperatif gözlemde, posterior cul de sak, rektum serozası ve sağ pelvik yan duvarda ince cidarlı, multikistik pelvik kitle saptandı. İntraoperatif patoloji konsültasyonunda kitle için ilk izlenim malign hücrelerin izlenmediği şeklindeydi. Pelvik kitlenin rezeksiyonuyla birlikte total abdominal histerektomi ve bilateral salpigo-ooforektomi yapıldı. Nihai patoloji sonucu BMPM olarak rapor edildi.

Sonuç olarak, BMPM nadir, benign kistik bir tümördür ve çoğunlukla preoperatif dönemde over kanseri olarak değerlendirilmektedir. İntraoperatif bulgular ve görünüm kanseri taklit edebilir. Bu nedenle frozen/section gereksiz agresif tedaviyi önleyecektir.

Anahtar Kelimeler: Benign multikistik peritoneal mezetelyoma, Peritoneal kist.

References

1. 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
2. O'Neil JD, Ros PR, Storm BL, Buck JL, Wilkinson EJ. Cystic mesothelioma of the peritoneum. Radiology 1989; 170;333-7
3. Szollosi A, Ferenc C, Pinter T, Erenyi A, Nagy A. Benign cystic mesothelioma, a rare tumor of the peritoneum. Magy Seb 2005;58:35-7
4. Ross MJ, Welch WR, Scully RE. Multilocular peritoneal inclusion cysts (so-called cystic mesotheliomas). Cancer 1989; 64: 1336-46
5. Tao LC. Aspiration biopsy cytology of mesothelioma. Diagn Cytopathol 1989;5:14-21
6. Bhandarkar DS, Smith VJ, Evans DA, Taylor TV. Benign cystic peritoneal mesothelioma. J Clin Pathol 1993;46; 867-8
7. Carpenter HA, Lancaster JR, Lee RA. Multilocular cysts of the peritoneum. Mayo Clin Proc. 1982 ;57(10);634-8
8. Letterie GS, Yon JL. The antiestrogen tamoxifen in the treatment of recurrent benign cystic mesothelioma. Gynecol Oncol 1998;70;131-3
9. Letterie GS, Yon JL. Use of a long-acting GnRH agonist for benign cystic mesothelioma. Obstet Gynecol 1995;85: 901-3