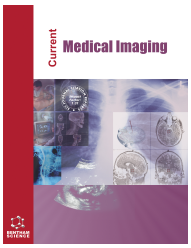
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CASE REPORT

Benign Multicystic Peritoneal Mesothelioma: Two Rare Cases and Review of the Literature

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Abstract:

Background:

Benign multicystic peritoneal mesothelioma is a multiloculated cystic mass which originates from the peritoneum. This rare tumor is usually seen in women of childbearing age and has a high recurrence rate after surgery.

Case Presentation:

We present two benign multicystic peritoneal mesothelioma cases with different imaging modalities, which were also pathologically proven.

Conclusion:

The imaging features which may be diagnostic should be well known as there are very few reports regarding this entity.

Keywords: Benign multicystic peritoneal mesothelioma, Multicystic mass, Cystic mesothelioma, Peritoneal tumor, Magnetic resonance imaging, Ultrasonography, Computed tomography.

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1. INTRODUCTION

There are plenty of peritoneal neoplastic and non-neoplastic lesions. Benign multicystic peritoneal mesothelioma (BMPM) is a subtype of mesothelial tumor which presents as a slow growing multi-septated cystic mass and has little risk for malignant transformation [1]. Main treatment of this rare entity is cytoreductive surgery with hyperthermic intraperitoneal chemotherapy (HIPEC), although a high recurrence rate is expected [2, 3]. Clinical findings are generally not specific and depend on the location of the tumor and the presence or absence of compression on adjacent organs. Since there are only a few reports about this entity, one should be familiar with its radiologic features. For this purpose, we are willing to share various appearances of BMPM with different imaging modalities.

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2. CASE REPORTS

2.1. Case 1

A 38-year-old male patient was admitted to our hospital with intermittent mild abdominal pain that started two months ago. Physical examination revealed a palpable mass in the right lower quadrant and umblical region. His medical history was unremarkable other than prior appendectomy. Complete blood count and routine biochemistry showed no abnormality. An ultrasound examination was planned. Grey-scale ultrasound images demonstrated the multi-septated cystic mass (Fig. **1**). Ultrasound guided aspiration was performed and 12 ml sample was taken for biochemical and microscobic evaluation (Fig. **2**). Laboratory evaluation of the aspirate showed normal creatinine, cholesterol, and bilirubin levels, but significantly increased cancer antigen 125 (CA-125) by 105440 units/mL (normal range is 0-35 units/mL). Microscopic specimen evaluation was negative for the protoscolexia of *Echinococcus granulosus*. Following these, computed tomography (CT) and magnetic resonance imaging (MRI) were performed. CT

showed the hypoattenuating multicystic mass and confirmed the ultrasound findings (Fig. **3**). MRI showed that the lesion originated from the subhepatic area on the right along the ascending colon and small intestine segments and moved to the left of the midline at the umbilicus (Fig. **4**).

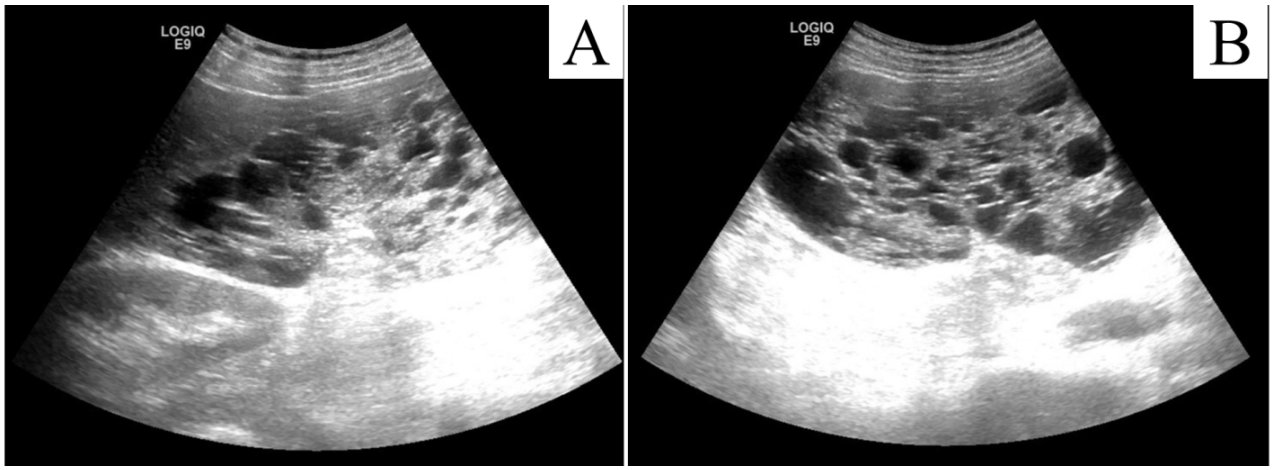


Fig. (1). Grey-scale ultrasound examination shows multiseptated cystic mass in subhepatic (**A**) and pelvic (**B**) region.

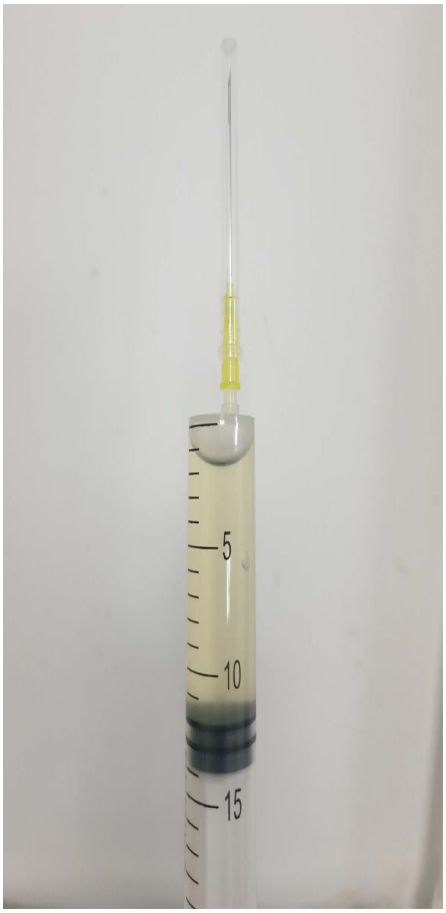


Fig. (2). The aspirate is clear and serous in appearance.

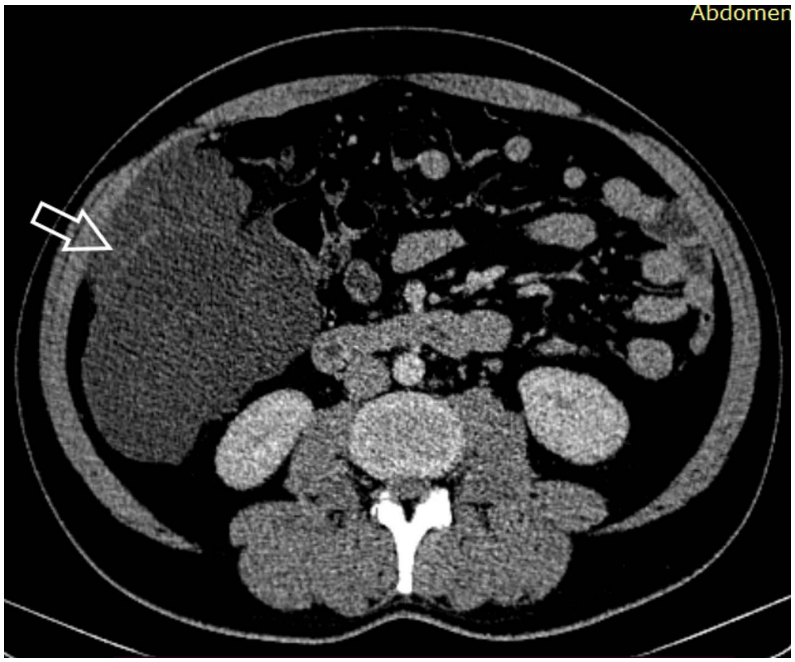


Fig. (3). Axial contrast enhanced CT slice demonstrates low density lobulated mass (arrow) with noticeable septa formations.

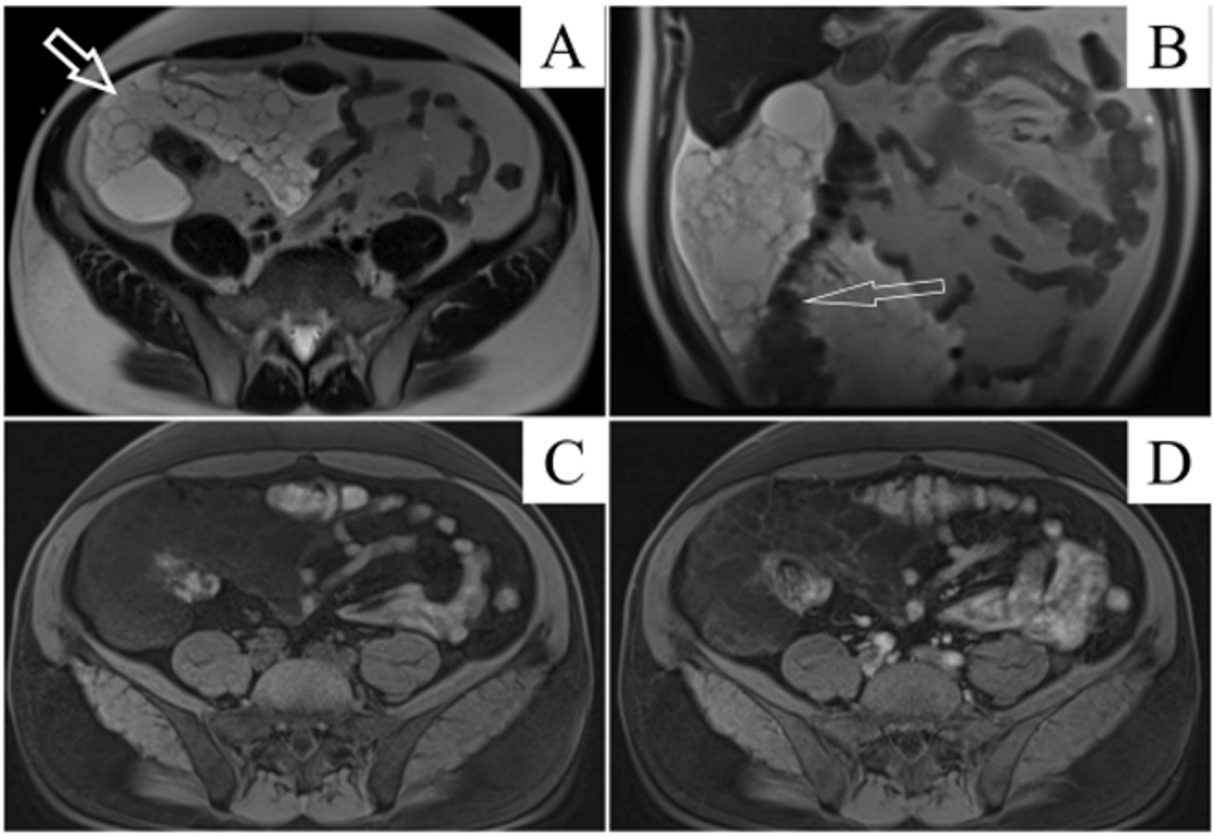


Fig. (4). (**A**) Axial T2W MRI shows hyperintense complex cystic lesion with thin septations (thick arrow). The mass crosses the midline . (**B**) Coronal T2W images demonstrates extension of the lesion to the pelvis adjacent to the ascending colon (thin arrow) more clearly. Pre (**C**) and post (**D**) contrast T1W slices shows the enhancement of septa formations (arrows).

The patient was operated and the cystic mass lesion was removed. On macroscopic and microscopic examination, the lesion appears to be of a cystic nature (Fig. **5**). On immunohistochemical examination, the lesions show diffuse nuclear and cytoplasmic staining for WT-1 and Cytokeratin 7 (Fig. **6**).

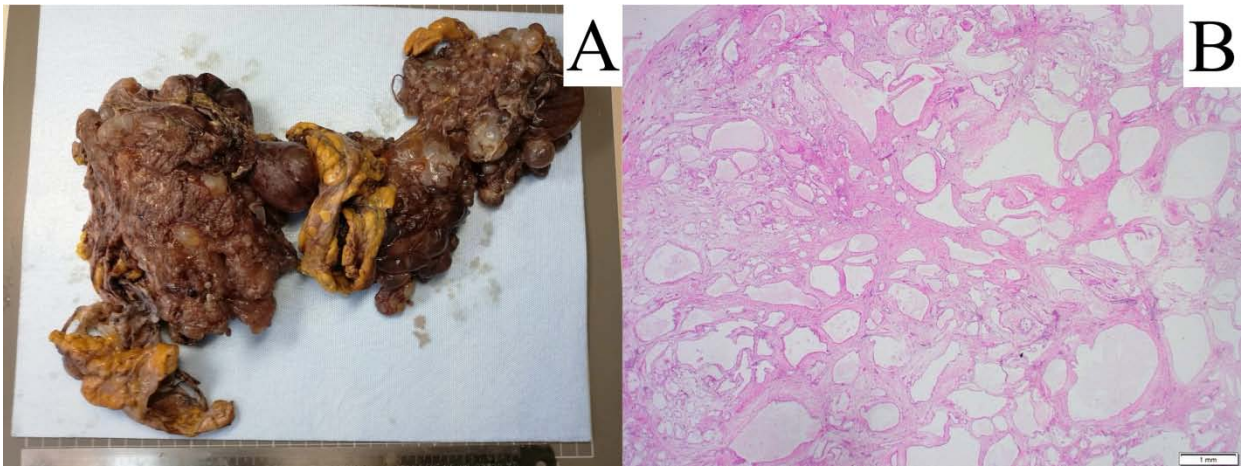


Fig. (5). (**A**) Material consists almost entirely of cystic lesion filled with serous fluid, intermingled with neighbouring adipose tissue. (**B**) Multiple cystic spaces of varying shapes and sizes are easily seen in a myxoic hypocellulary background.

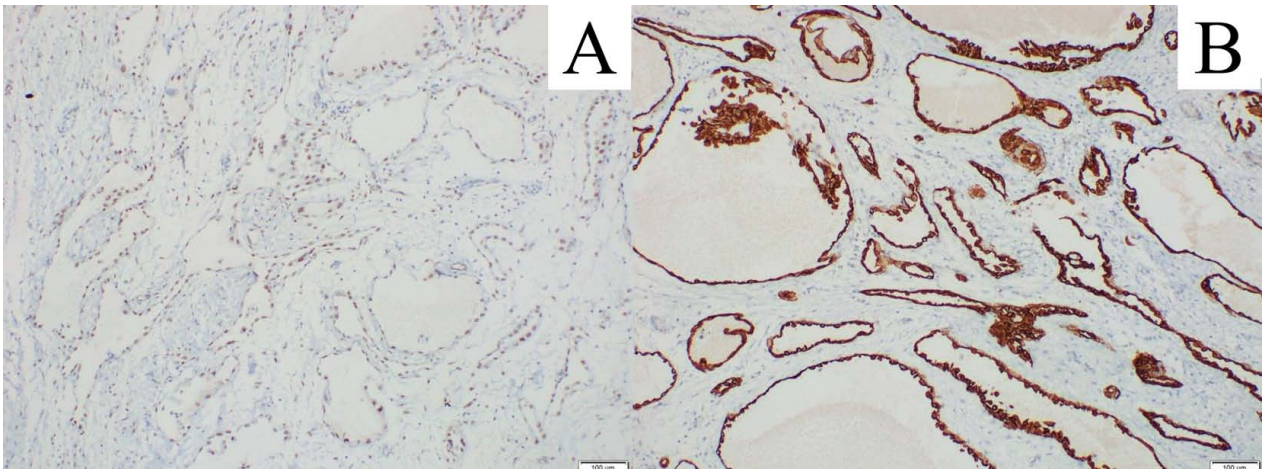


Fig. (6). (**A**) WT-1 immunohistochemistry (100x). Diffuse, moderate nuclear staining in tumor cells. (**B**) Cytokeratin 7 immunohistochemistry (100x). Diffuse strong cytoplasmic staining in tumor cells.

2.2. Case 2

A 59 year old male patient presented with the complaint of long-standing epigastric pain. No findings were found in the physical examination and the patient had no previous history of surgical operation. Complete blood count and routine biochemistry tests showed normal results. Endoscopy was performed with the pre-diagnosis of gastritis. Cross-sectional imaging was planned for the patient who had signs of external compression on the gastric corpus on endoscopy. CT and MRI were also taken. On CT, a cystic lesion was seen starting from the lower part of the spleen and ending in the left paracolic region at the level of the umbilicus (Fig. **7**). MRI demonstrated the well-defined multiseptated cystic mass in the perisplenic region (Fig. **8**). The patient then underwent surgery at another hospital. In the pathological examination of the material, cystic structures containing a large number of serous transparent fluids were seen on the pathological tissue and the diagnosis was confirmed.

3. DISCUSSION

There are many peritoneum based neoplastic lesions but very few cases of BMPM have been published to date. Its mesothelial provenance was first defined by Mennemeyer and Smith in 1979 by using electron microscopy [4]. Pathophysiologically development of BMPM is not well known yet, but some authors link it with previous abdominal surgery, endometriosis, pelvic inflammatory disease and recurrent peritoneal inflammation [5]. Unlike malignant mesothelioma, there is no proof in the literature that suggests asbestos exposure is related to BMPM [6, 7].

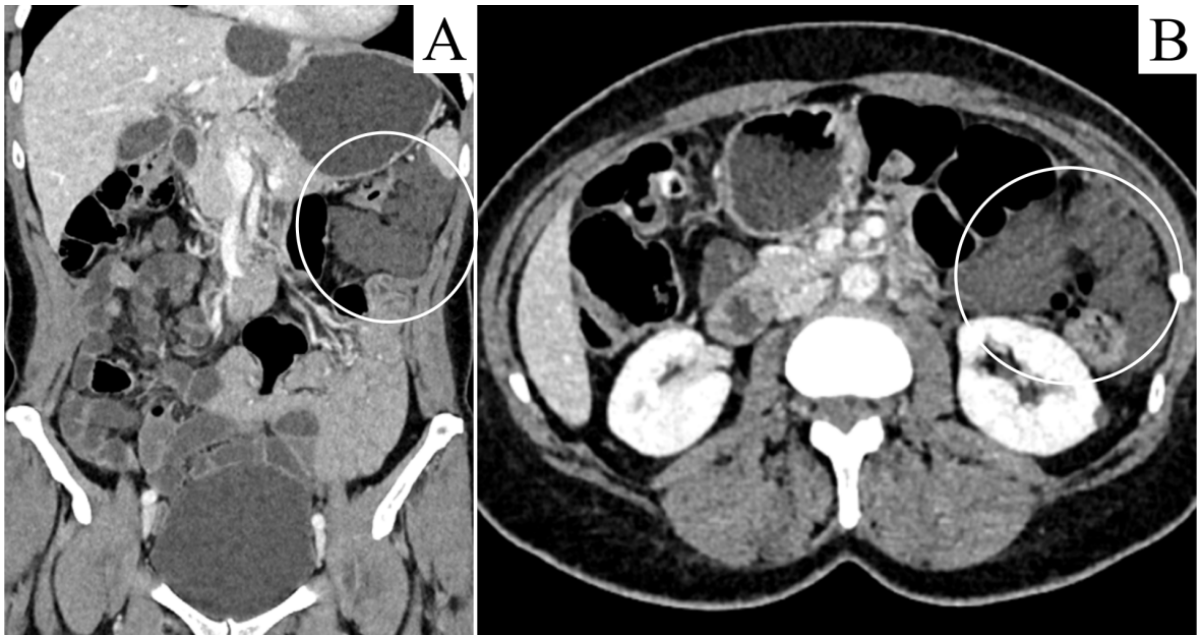


Fig. (7). Coronal (**A**) and axial (**B**) CT images shows the hypoattenuating mass in the left upper quadrant.

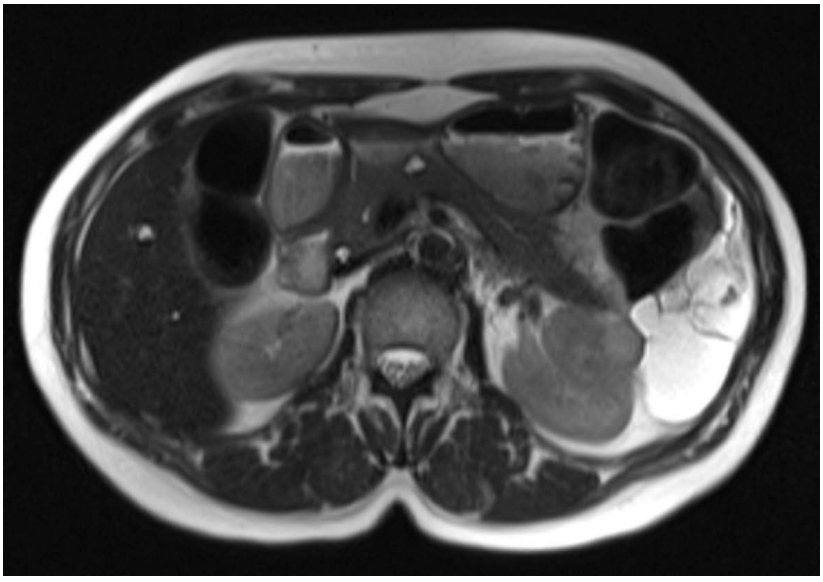


Fig. (8). Axial T2W MRI demonstrates the T2 hyperintense, fluid filled multicystic mass with septa formations.

In the pathologic examination, it is composed of variable sized cystic spaces surrounded by mesothelial cells. Immunohistochemical stains exhibit reactivity for calretinin, vimentin, ceratin, CA-125 and uncommonly for estrogen receptor and progesterone receptor [8]. Although these cysts usually contain serous fluid, there may be occasionally hemorrhagic and gelatinous content [9].

In the literature, it is stated that it is more appropriate to define multicystic peritoneal mesothelioma as a low-grade or borderline tumor, rather than the benign category, because of the high risk of recurrence and rarely, malignant transformation [1, 10].

In imaging, the radiography may show a soft tissue density structure that pushes the adjacent intestinal loops and occupies space in the abdomen [11]. However, ultrasound is often used as the first step in imaging and shows a multicystic lesion with septation. Color doppler shows no flow in the mass. CT and MRI can better show the extent of the lesion. The cysts are hypoattenuating in computed tomography [12]. MRI can show T1 hypointense and T2 hyperintense cystic lesions [13]. Although the internal components of the cysts do not show enhancement after contrast injection, septa formations may enhance. There are also cases in the literature with intraabdominal free fluid and hemorrhage [11].

In comparing our cases with those of the literature, the imaging characteristics were similar. Although the walls of the cysts can be enhanced, there was no significant nodular enhancing component either in our cases or in the literature.

There was no calcification in the cyst walls or septa in our case although calcification has been reported in the cyst walls or septa in some cases [11 - 13].

Cyst contents in T1-weighted sequence were generally hypointense in the literature as in our case. However, in cases of intracystic hemorrhage, the cyst content may be T1W hyperintense. Invasion of the multicystic mass into adjacent organs and structures has not been demonstrated so far. Intracystic fat has not yet been documented [14].

The clinical findings are generally vague as long as this tumor does not cause compression symptoms in adjacent organs, it usually does not cause clinical symptoms. A wide range of clinical conditions from pneumoperitoneum to urinary incontinence have been described in the literature [15, 16]. The involvement is usually in the pelvis, although it can be anywhere on the peritoneal surface [17].

In the literature, BMPM cases developing from the pleura, pericardium and tunica vaginalis have also been reported [18 - 20]. Studies in the literature have also shown that in the abdomen, BMPM is more common in the right half than in the left [14].

Cystic lmpyhangioma is the major differential diagnosis of BMPM. However, cystic lymphangioma is usually seen at younger ages and there is no significant difference in distribution between the genders. Other differential diagnoses include cystic tumors of the ovary, endometriosis and pseudomyxoma peritonei [21]. In the case of pseudomyxoma peritonei, there is often a mucinous tumor of appendix origin and diffuse low-density mucinous implants “scallop” the margins of the visceral serosal surfaces [22].

Although radiological imaging narrows the differential diagnosis to a great extent, it is sometimes insufficient and patients are diagnosed with pathological examination after the operation.

Various treatment strategies have been used, including surgical excision, hormonal therapy, sclerotherapy and laser vaporisation. Today, cytoreductive surgery and HIPEC come to the fore in the surgical treatment of this tumor. Cisplatin and doxorubicine are usually used in the intraoperative chemotherapy protocol [1, 3]. The prognosis is good as malignant transformation is usually rare [17].

CONCLUSION

Because BMPM is a very rare tumor and clinical symptoms are often unclear, radiology is important in the diagnosis. Since the number of cases described in the literature are rare, we think that the imaging findings in different radiological modalities in our study will contribute to the literature.

AUTHORS' CONTRIBUTION

M.M.I., M.U. and G.E. conceived, designed, and organized the study. R.K. and B.S. performed the interpretation of data, M.M.I. wrote the manuscript. All the authors read and approved the final manuscript.

LIST OF ABBREVIATIONS

BMPM	=	Benign Multicystic Peritoneal Mesothelioma
HIPEC	=	Hyperthermic Intraperitoneal Chemotherapy
CT	=	Computed Tomography
MRI	=	Magnetic Resonance Imaging

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

Not applicable.

HUMAN AND ANIMAL RIGHTS

Not applicable.

CONSENT FOR PUBLICATION:

Informed consent was obtained from the participants included in the study.

STANDARDS OF REPORTING

CARE guidelines were followed.

AVAILABILITY OF DATA AND MATERIALS

The data and supportive information are available within the article.

FUNDING

No funding was received for this study.

CONFLICT OF INTEREST

On behalf of all authors, the corresponding author states that there is no conflict of interest.

ACKNOWLEDGEMENTS

Declared none.

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