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Challenges of Benign Multicystic Mesothelioma: Recurrence and Metastasis- A Rare Case Report

Tarik Alam Ony[1], Anjuman Sultana[2], Mohammad Haroon Or-Rashid[3], Mir Rasekh Alam Ovi[4] and Md. Nashir Uddin[5]*

[1]*Department of General Surgery, Mackay Base Hospital, Dhaka, Bangladesh*

[2]*FCPS (Obstetrics & Gynaecology), Mugda Medical College & Hospital, Dhaka, Bangladesh*

[3]*FCPS, MRCSEd, MS (Colorectal Surgery), Dhaka Medical College & Hospital, Bangladesh*

[4]*Department of Surgery, Dhaka Medical College & Hospital, Dhaka, Bangladesh*

[5]*FCPS (Surgery), MRCS (Glasgow), Department of Surgery, Mugda Medical College Hospital, Dhaka, Bangladesh*

Abstract

A rare condition called multicystic mesothelioma, also known as benign multicystic peritoneal mesothelioma (BMCM), originates from parietal or visceral peritoneum mostly affecting females of reproductive age, often challenging to diagnose accurately prior surgery. We present a 28-year-old woman with a image guided core biopsy proven mucinous ovarian adenocarcinoma who underwent definitive surgery, her biopsy surprisingly revealed benign multicystic mesothelioma. Since multicystic mesothelioma can behave like locally malignant tumors despite its benign origin, an accurate diagnosis is crucial to formulate appropriate management plan. This case report highlights the need for awareness of multicystic mesothelioma due to its rarity, diagnostic complexity, and potential for recurrence despite its generally benign behavior. Further research is warranted to elucidate the pathogenesis, optimize treatment strategies, and improve patient outcomes. Comprehensive diagnostic and surgical strategy play a crucial role in this regard.

Keywords: Benign; Multicystic; Mesothelioma; Recurrence; Metastasis

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Correspondence:
Anjuman Sultana, FCPS (Obstetrics & Gynaecology), MRCOG Mugda Medical College & Hospital, Dhaka, Bangladesh, E-mail: anjumansultana@gmail.com

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Introduction

Cystic mesothelioma is a sporadic condition that commonly arises from the peritoneal mesothelium, but it can also occur in other areas such as the pleura, pericardium, tunica vaginalis, and spermatic cord [1-3]. In peritoneal cavity it can be found attached to various organs such as the intestine, omentum, spleen, liver, or retroperitoneal space, and it rarely occurs in extra-abdominal locations. Multicystic mesothelioma is typically diffuse and tends to develop on the surfaces of pelvic organs. Though this tumor is benign in nature, it often has a high rate of local recurrence. BMCM is five times more common in female compared to male, and a few cases have also been reported in children. Clinical presentation, diagnostic imaging, and operative findings of BMCM are often similar to those of ovarian carcinomas [1-3].

There is controversy regarding the pathogenesis of multicystic mesothelioma, with some researcher reported a neoplastic process while others suggest a reactive process [2,3]. Many patients with multicystic mesothelioma gave a history of intrapelvic surgery for endometriosis, uterine leiomyoma, or inflammation. A hypothesis is postulated that this tumor is the result of peritoneal reaction to chronic irritation with mesothelial cell entrapment followed by reactive proliferation, and cyst formation. BMCM appears histologically as cystic spaces lined by low cuboidal cells with hobnail features with vascularity [2,4].

The clinical presentation of multicystic mesothelioma is generally nonspecific. Patients often present with unclear vague abdominal pain and distention, palpable masses in the abdomen, dyspepsia, ascities [3-5]. It may also be discovered incidentally during routine diagnostic tests, general medical examinations, or surgical procedures. Differential diagnoses for this condition include cystic lymphangioma, mucinous cystadenoma, cystic teratoma, and pseudomyxoma peritonei. Treatment modalities range from conservative management to complete resection, followed by hyperthermic intraperitoneal chemotherapy (HIPEC). However, there is currently no consensus on the standard treatment and follow-up of patients with this condition [4,5].

Case Presentation

A 26 yr old nulliparous regular menstruating female got admitted into Bangabandhu Sheikh Mujib Medical University with chief complaints of abdominal distension with feeling of heaviness in her lower abdomen for 2 months. Her pre-morbid status was fairly unremarkable without any significant medical, surgical or gynecological history. She did not describe any weight loss and change in bowel habit.

On presentation, her vitals were all within normal limit. Her abdominal and pelvic examination revealed a 15 cm × 15 cm, non-tender, mixed solid-cystic mass with irregular surface and border, occupying predominantly umbilical, hypogastric, right lumber and right iliac fossa. Bed side examination revealed positive shifting dullness raising suspicion of ascites.

Ultrasound showed a fairly large 17 cm^2 × 16.5 cm^2 mass having both solid and cystic components, occupying pelvic cavity with moderate ascites. CT scan appreciated a ovarian malignant looking mass 18 cm^2 × 16.5 cm^2 involving left ovary, with mesenteric and omental seeding, para-aortic lymphadenopathy and moderate ascites, giving primary impression of an advanced stage of ovarian cancer.

Her Ca-125 was elevated to 1034iu/ml but Ca19-9, LDH, Alfa feto- protein were within normal range. She had normal endoscopy and colonoscopic workup. Ascitic fluid study reported no malignant cell. Ultrasound guided core biopsy from ovarian mass showed malignant tumour made of epithelium cells present in cord and tubules, stroma with myxoid areas where pathologist remarked as Mucinous Cystoadenocarcinoma (Figure 1).

She was discussed in tumor board and was planned for neo-adjuvant chemotherapy followed by interval debulking surgery. However, her tumor response was not satisfactory after 3 cycles of chemo with cisplatin and paclitaxel, therefore, she was taken to theatre.

Operative findings were -hemorrhagic peritoneal fluid on entry into peritoneal cavity. Loosely attached multiple cystic lesions of variable sizes involving entire parietal peritoneum, pouch of doglas, bladder serosa and undersurface of diaphragm. A 17 cm × 16 cm angry looking mass was noted in attached with left Fallopian tube which was impacted in pouch of Douglas however, the mass was separated from left ovary. Both ovaries were with metastatic deposits, omentum was turned into cake. On given findings, a total abdominal hysterectomy, bilateral salpingo-oopherectomy with total omentectomy and removal of metastatic deposits were done.

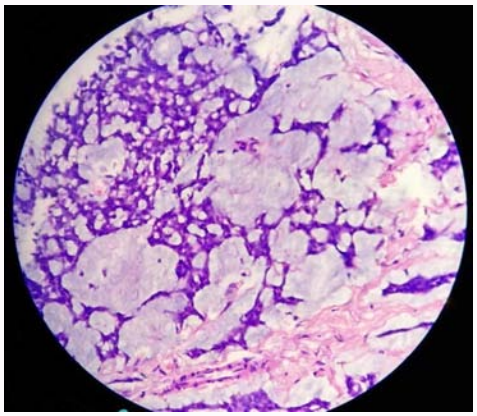

Figure 1: Biopsy report following first core biopsy from left adnexal mass showing tumor made of epithelium cells present in cord and tubules, stroma with myxoid areas suggestive Mucinous Cystoadenocarcinoma.

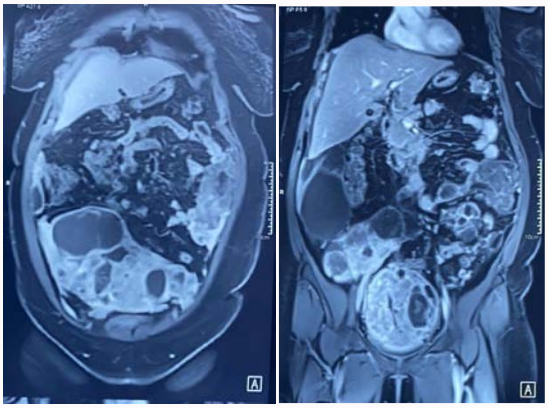

Figure 2: MRI of whole abdomen, (a,b-coronal view) showing a complex mass with intermediate solid and hypointense cystic component with septation measuring 23.5cm x 25 cm following interval debulking surgery.

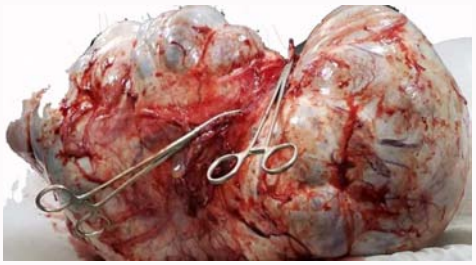

Figure 3: Photograph showing intrabdominal mass of secondary debulking surgery.

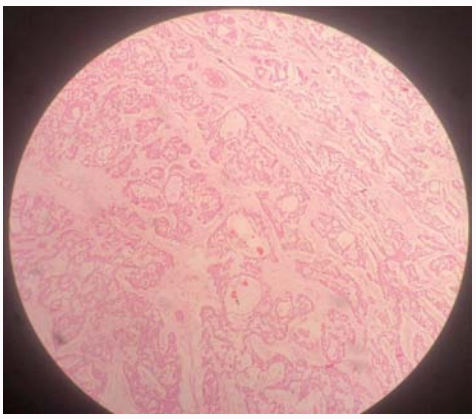

Figure 4: Histopathology report of specimen of secondary debulking surgery consisting tissue from tumour mass showing atypical cuboidal cells having prominent nucleoli with eosinophilic cytoplasm arranged in tubules micropapillae and trabeculae. Area of myxoid stroma and cystic changes with infrequent mitosis were seen.

Histopathology reported cuboidal cells with mild nuclear pleomorphism and cells were arranged in clusters floated in extra cellular mucin. Mitosis was infrequent and there was no evidence of stromal involvement, lymphovascular stromal invasion and malignancy. Tumour deposited on ovarian capsule but stroma was free from tumour. Immunohistochemistry showed calcitonin, WT-1, AE3, AE1, CD 34 positive in tumour cells while Desmin, podoplanin

were negative. Final diagnosis was benign cystic mesothelioma.

She had unevenful recovery and remained symptom free for 2 years before representing again with abdominal distension. In her re-presentation, clinically she was again found to have larger sized nearly 20 cm × 25 cm mass in abdomen with no ascites. MRI revealed a solid-cystic complex mass measuring 23.5 cm × 25 cm in the abdomen, with the remaining intraperitoneal organs appearing normal (Figure 2). Ultrasound guided core biopsy was performed again which reported mucinous cystoadenocarcinoma. CA 125 was elevated to 801 iu/ml.

She was planned for upfront secondary debulking surgery revealing a 25 cm x 22.5 cm complex mass in abdomen (Figure 3), multiple grape like cystic structure freely attached with peritoneum, intestine, spleen, undersurface of liver, pancreas. Histopathology returned again as cystic mesothelioma with IHC similar findings (Figure 4). Six months later the patient again developed abdominal distension. She was re-discussed in tumor board and was planned to have surgical surveillance. She declined the option for re-operation and six months later she passed away.

Discussion

Around 150 instances of multicystic mesothelioma have been documented in medical literature since Mennemeyer and Smith first reported it in 1979. Multicystic mesothelioma appears to primarily affect women of reproductive age, who have history of asbestos exposure, prior abdominopelvic surgeries, pelvic inflammatory disease or endometriosis. About 80% of cases occur in women, typically those in their late twenties to mid-thirties (with an average age of 37). This patient described here belonged to reproductive age group but mentioned no such risk factors as well as family history [4-6].

Mesothelioma rarely affects the ovaries, with ovarian mesotheliomas accounting for only 0.03% of mesothelioma-related deaths in the United Kingdom registry over a period of 24 years. Common symptoms include abdominal pain, swelling, and sensitivity, frequently accompanied by nausea and vomiting, and rarely ascites. The cases we encountered exhibited the above symptoms along with ascites, which made think of ovarian malignancy due to the similarity in clinical features [6,7].

Multicystic masses in BMCM are detected by ultrasound, CT scan or MRI. All imaging techniques may reveal free-fluid in the peritoneal cavity with minimal peritoneal thickening, which is in contrast to intra-abdominal malignancy involving parietal peritoneum, where there is often frank ascites, diffuse peritoneal thickening, organ metastasis. Diagnosis is achieved through surgical sampling with immunohistopathologic studies. It has also been suggested that patients with high levels of CA125 may have worse biological behavior of the disease. Our patient also had high level of CA125 and ultimately, she developed recurrence and poor outcome [7,8].

When mesotheliomas occur in the ovary, it is important to distinguish them from other neoplasms such as cystic lymphangioma, cystic forms of endosalpingiosis, endometriosis, and cystic adenomatoid tumors [3,6]. A small number of cases with mesothelial tumors were initially misdiagnosed as ovarian tumors before surgery. In this case, the presence of mucinous cystoadenocarcinoma in the core biopsy also created a misdiagnosis for which our patient received three cycle chemotherapy. Immunohistochemistry was needed to reach the conclusion [4,7,9].

Under the microscope, these cysts are covered by tubal epithelium, which includes varying proportions of ciliated cells, nonciliated secretory cells, and sometimes intercalated cells. The international mesothelioma interest group identified calretinin, cytokeratin 5/6, WT-1 protein, podoplanin, mesothelin. The immune histochemistry results of this case also shows WT-1, positive but podoplanin negative [3,8].

Complete surgical removal is the most effective treatment for BMCM, but there is a high rate of local recurrence, ranging from 33% to 50%. Chemotherapy and radiotherapy have not been found to be effective in treating multicystic mesothelioma [1,3].

Although multicystic mesothelioma is typically benign, recurrence is more common in cases with large masses or extensive disease, as seen in our case. Despite the histologically benign nature of the disease in this patient, both recurrence and metastasis have been observed. Metastasis can be explained by direct and transcoelomic spread to distant organs, as also noted in other literature [4,8]. The benign nature of the disease is histologically evidenced by mild nuclear pleomorphism, infrequent mitotic figures, and the absence of stromal invasion. In contrast, malignant tumors exhibit pronounced nuclear atypia, frequent mitosis, and lymphovascular stromal invasion. Malignant transformation of multicystic mesothelioma has been reported in two cases [6,7,9].

Conclusion

The difficulty in preoperative or intraoperative diagnosis of multicystic mesothelioma has been demonstrated, particularly in distinguishing it from an ovarian malignancy for which this patient received chemotherapy and remained non responsive. Additionally, the rarity of this disease, presence of metastasis & recurrence in spite of being benign makes it challenging to manage appropriately. Advancing novel diagnostic markers for multicystic mesothelioma is crucial for future clinical practice.

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Approval of the Research Protocol: The permission for case report was approved by Institutional Review Board of Bangabandhu Sheikh Mujib Medical University, Dhaka Bangladesh. BSMMU/2022/686, date 21-12-2022.

Informed written consent: Informed written consent was obtained from patient for writing and publishing this case report.

Orchid ID: Anjuman Sultana. orcid.org/0000-0001-6601-4999

Data Availability Statement

Data sharing is not related to this article, as no new data were

generated or analyzed during this study.

Author Contributions

1. Development of idea, drafting, editing & finalization of article.
2. Management, coordination and supervision of research activity and finalization of article.
3. Reviewing & editing of article, literature review & draft preparation.
4. Literature review and drafting the article.
5. Literature review.

All co-authors have seen and agreed with the contents of manuscript.

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