Benign Pleural Multicystic Mesothelioma: A Rare Case Report

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Case report

Keywords: Multicystic mesothelioma, Pleura, Lung

Posted Date: February 2nd, 2021

DOI: https://doi.org/10.21203/rs.3.rs-165790/v1

Abstract

Background: Fewer than 200 benign multicystic peritoneal mesothelioma cases were reported worldwide till 2017, while its pleural involvement has rarely been reported.

Case presentation: We report a 70-year-old man who presented with three months history of chronic cough. Surgical resection was performed, and the pathology confirmed benign multicystic pleural mesothelioma. The patient underwent right lateral thoracotomy, wedges resection of the right upper lobe, and parietal pleurectomy and was discharged with an uneventful postop course.

Conclusion: Based on published literature to date, this is the second reported case of pleural involvement of this disease.

1. Introduction

Multicystic benign mesothelioma (MBM) is a rare benign tumor that presents with recurrent mesothelial cysts that arise from the epithelial and mesenchymal elements of mesothelial tissue. It has a meager potential of transformation to malignant mesothelioma. [(1)] BMPM was first described in 1928 by Plaut, who incidentally observed loose pelvic cysts during an operation for uterine leiomyoma. The mesothelial origin was later confirmed by electron microscopy. [(1)] This cystic structure usually arises from peritoneal tissue and mostly involve female in reproductive age, rare cases have been described in men. [(2)] The etiology of the disease is ill-understood. Some consider it to be a reactive process secondary to previous surgical trauma or inflammation cause. [(2)] We present a very rare case of MBM in the pleural cavity of a 70 years old male. To our knowledge, it is the second case of multicystic benign pleural mesothelioma till the date of this report.

2. Case Report

A 70 years old male, presented with chronic dry cough for three months and a past medical history of renal stone and benign prostatic hyperplasia (BPH), referred to our clinic. Negative history of any thoracic surgery or other underlying health conditions. Inflammatory blood markers such as erythrocyte sedimentation rate and C-reactive protein were increased. His laboratory results were normal except; low hemoglobin level and increased prostatic specific antigen level.

The abdominopelvic ultrasonography report indicated a 31 mm cortical cyst in the right kidney, mild pleural effusion on the right side, and enlarged prostate (43 ccs) with a homogenous echo pattern. The CT imaging without contrast showed a focal grand-glass opacity (crazy paving appearance) at the apex of the right lung, at least five sub-pleural, mediastinal base pulmonary nodules with a maximum size of 9 mm, and a pulmonary nodule of 18 mm*16 mm at the right lower lobe of the lung.

Fine nodular aspiration (FNA) was performed on right pulmonary nodules. The microscopic evaluation of FNA results showed cellular smears composed of many isolated groups, a cluster of atypical epithelial

cells, and macrophages in a bloody background with high malignancy

suspicion.

A chest CT scan with IV contrast was performed for further evaluations, which showed a 29 mm pleural base irregular nodule in the apex of the right lung with superior soft tissue extension without significant ribs or vascular invasion. Also, an 18 mm nodule in the base of the right lung base, multiple pleural base nodules up to 12mm, was seen in the right lung, highly suspicious of a metastatic lesion. Based on these findings, the patient was scheduled for surgery.

After prepping and draping, under general anesthesia, the skin was incised through a standard lateral thoracotomy incision. The pleural cavity was entered through the 4th intercostal space with adhesion presenting between parietal and visceral pleura, and partial parietal pleurectomy was done, then the pleural cavity was evaluated. A mass lesion was present in the right upper lobe; therefore, wedge resection was performed on the right upper lobe, followed by parietal pleurectomy. The tissues were sent to the pathologist, and the patient was discharged with an uneventful post operation course.

Based on the lesion's pathological evaluation, the gross evaluation showed multiple irregular fragments of creamy- gray and yellow rubbery tissue, measuring (5*4*1.3) cm, demonstrating multiple benign lesions, consistent with benign multicystic mesothelioma. Immunohistochemistry (IHC) study was performed to confirm the diagnosis and roll out other differential diagnoses such as alveolar adenoma and sclerosing hemangioma, which demonstrated CK7 positive, CD34 negative, Calretinin positive, TTF1 negative, EMA weak, P53 negative; which were in favor of "multicystic mesothelioma."

Three months after surgery, a follow-up spiral chest CT scan was done, demonstrating evidence of loculated pleural effusion at the right lower lobe periphery. After injection of contrast, evidence of small wall enhancement with the possibility of empyema formation was diagnosed. Also, there were multiple enhancing nodules in the pleural on the right side scattered in both upper and lower zones of the chest's right side. According to this finding possibility of multicystic mesotheliomas was considered. Also, a mass lesion noted at the right apex of the lung with some heterogeneous enhancement measuring 3*2 cm in size seems to be mesothelioma's focuses. Multiple similar nodularities in the pleural cavity's supradiaphragmatic area were noted due to multiple nodules of mesothelioma.

The specimens from our patient's lesion were obtained and sent to the pathology lab, which demonstrated multiple cysts and cystic nature of this lesion with thin walls, lined with one layer of mesothelial cells, which has flattened to cuboidal morphology (Fig. 1), which these findings were in favor of multicystic mesothelioma.

3. Discussion

Multicystic benign mesothelioma (MBM) is a rare benign neoplasm associated with no well-defined symptoms, typical clinical or imaging data. [(3)] Over the last decade, the most common treatment has

consisted of complete resection of the tumor. However, after resection, the recurrence rate is approximately 50% after a period of 3 to 27 months (mean 32 months) [3]. Classically these tumors present as large multicystic masses with thin-walled septations. [4]

Diagnosis is achieved through surgical sampling with histopathological examination. Immunobiologically, BMPM exhibits multiple small cystic spaces with flattened lining containing calretinin positive cells without atypical features, mitotic figures, or tissue invasion. Treatment includes cytoreductive surgery. [5] MBM is a rare disorder that occurs predominantly in reproductive-aged women. [6] There are few case reports of this condition in males. [7] A ratio of 5 women:1 men has been reported. [7] Most MBM cases involve the peritoneum and abdominal cavity, and some cases have been reported involving vaginal tunica. [7] MBM is commonly asymptomatic and often an incidental finding on physical, imaging, or intraoperative and can affect organs by contiguity. [7] Our report is a rare case of a male patient with a diagnosis of MBM of pleura. There is only one case report of multicystic benign pleural mesothelioma to our knowledge. [8]

Due to this condition's high recurrence rate, we followed-up with the patient and performed a spiral chest CT scan three months after the surgery. Evidence of loculated pleural effusion at the periphery of the right lower lobe was seen in the CT scan then after contrast injection, small wall enhancement with the possibility of emphysema formation was diagnosed. The CT scan indicated; multiple enhancing nodules in the pleural at the right side scattered in both upper and lower zones of the right side of the chest. Also, a mass lesion was noted at the right apex of the lung with some heterogeneous enhancement measuring (3*2) cm, and it seems to focus on mesothelioma. Multiple similar nodularities in the supra-diaphragmatic area of the pleural cavity were also noted. According to this finding, the possibility of multicystic mesotheliomas was considered.

As indicated in other multicystic benign mesothelioma researches, treatment of multicystic benign mesothelioma is surgery and chemotherapy,(9) therefore one month after surgery, we referred our patient to an oncologist for chemotherapy treatment. In our case, if a sooner diagnosis was achieved along with aggressive surgical resection, there would not have been the need for chemotherapy since his condition was benign.

4. Declarations

Ethical approval of the study

Written inform consent was obtained from the patient in our study. The purpose of this research was completely explained to the patient and was assured that their information will be kept confidential by the researcher. The present study was approved by the Medical Ethics Committee of the academy.

Consent for publication

Consent was obtained from the patient regarding the publication of this case report.

Availability of data and materials

All data regarding this case has been reported in the manuscript. Please contact the corresponding author if you are interested in any further information.

Competing interests

The authors declare that they have no competing interests.

Funding

No financial support was received for this case report.

Authors’ contribution

AR made the disease diagnosis and carried out the patient's treatment course. RS and AE collected the data. AM carried out the pathological evaluation while BZ performed the surgery. MJF made a significant contribution in the patients' diagnosis and treatment course. SP drafted the manuscript. All authors proofread and accepted the final version of the manuscript.

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Figures

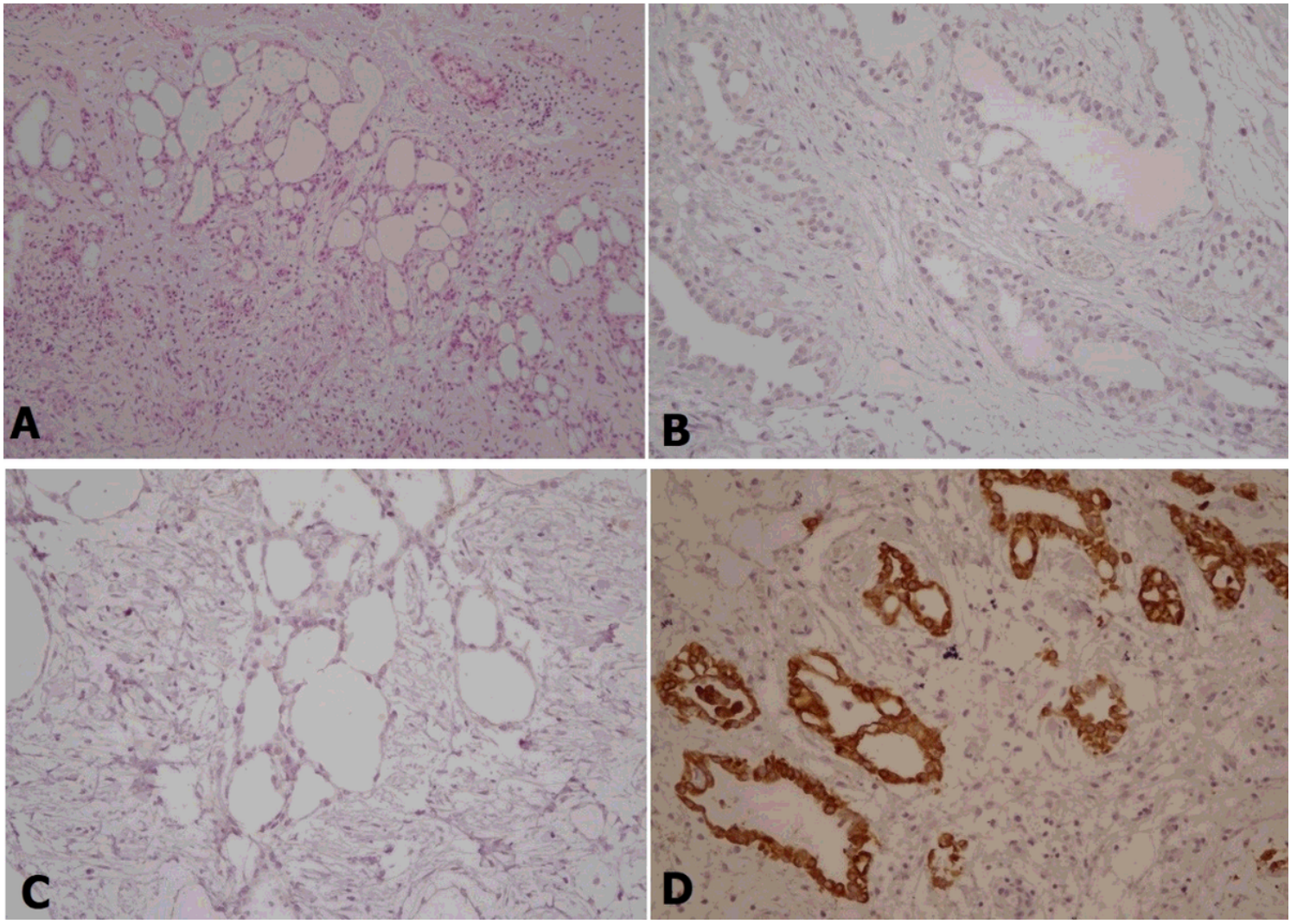

Figure 1

(A) H&E-Low power magnification shows multiple cysts filled with serous fluid.; (B) H&E-single layer of flattened to cuboidal mesothelial cells line cysts; (C) H&E-Lining of cysts composed of single layer mesothelial cell; (D) IHC- Calretinin positive cyst wall lining.