Case Report

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Challenging diagnosis of sarcomatoid hepatic mesothelioma: a case report with review of literature

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Introduction: Mesothelioma is a rare and aggressive cancer that is primarily caused by asbestos exposure. However, cases of mesothelioma without asbestos exposure suggest the involvement of other risk factors. Sarcomatoid mesothelioma, which is characterized by spindle-shaped cells, is a particularly aggressive subtype with limited treatment options.

Case presentation: The authors present a case of a 72-year-old man with no history of asbestos exposure who presented with abdominal pain, fatigue, and weight loss. Imaging revealed a large cystic mass in the liver. A Liver biopsy confirmed the diagnosis of sarcomatoid mesothelioma. Immunohistochemistry results further supported this diagnosis. Due to the advanced stage and tumor size, surgical resection was not feasible. Palliative chemotherapy was initiated, but the patient's condition deteriorated rapidly, leading to his demise.

Conclusion: This case highlights the complexity of mesothelioma and the need for further research to identify the nonasbestos-related risk factors. Understanding alternative causative agents and mechanisms is crucial for the early detection, the development of targeted therapies, and improving patient outcomes. The presented case contributes to the existing literature and aligns with the Surgical CAse REport (SCARE) Criteria.

Keywords: gastrointestinal, hepatic neoplasm, primary malignant mesothelioma, sarcomatoid mesothelioma

Introduction

Mesothelioma is a rare and aggressive type of cancer that affects mesothelial cells of the pleura, peritoneum, and pericardium[1]. It is widely recognized that asbestos exposure[2] is the leading cause of mesothelioma development, with studies which show 80% of all mesothelioma is due to asbestos exposure in their lifetime and establishing a strong link between the two. However, mesothelioma has been reported to occur in individuals without any known asbestos exposure, suggesting the involvement of other potential risk factors like family history, environmental factors, and lifestyle factors.

HIGHLIGHTS

- Sarcomatoid mesothelioma of the liver is a rare cause of a hepatic malignancy.
- On computed tomography, it looks like a heterogeneous mass with multiple cystic lesions.
- Complete tumor resection may be an effective way to treat the tumor.
- Diagnosis is the main challenge for the clinicians.

Sarcomatoid mesothelioma, characterized by spindle-shaped cells and a poor prognosis, is a particularly aggressive subtype of the disease[3]. This particular variant accounts for a minority of mesothelioma cases and is even scarcely reported in cases of hepatic malignancy, and it exhibits a particularly poor response to treatment options.

However, the incidence of this deadly form of cancer is low. The case of a 72-year-old man with sarcomatoid mesothelioma of the liver underscores the complexity of this disease and the need for further research to identify potential nonasbestos-related risk factors. Therefore, it is crucial for medical professionals and researchers to explore alternative causative agents and mechanisms to enhance early detection, develop targeted therapies, and improve patient outcomes.

Method

We report this case in line with the updated consensus-based Surgical Case Report (SCARE) 2020 criteria[4].

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Case presentation

We present the case of a 72-year-old man, who presented with a history of abdominal pain, fatigue, and weight loss of 10 kg over the past 6 months. He had a medical history of hypertension, diabetes mellitus, and coronary artery disease, but no history of tobacco or alcohol use. He did not report any history of asbestos exposure, elaborating the other environmental factors like carbon nanotube exposure, previous family history, or lifestyle factors all were negative.

On examination, he was found to have hepatomegaly, but no other significant findings. Abdominal imaging studies, including ultrasonography and computed tomography (CT), were performed. A CT scan showed a 13.6 cm × 11 cm mass in the liver with cystic liver (Fig. 1).

There was no evidence of metastasis to other organs. On a transverse CT scan, the cystic lesion occupying the right lobe is well appreciated (Fig. 2). A liver biopsy was performed, and a histopathological examination revealed spindle-shaped cells with pleomorphic nuclei and abundant cytoplasm, which was consistent with a diagnosis of sarcomatoid mesothelioma (Fig. 3).

Immunohistochemistry showed positive staining for cytokeratin, vimentin, calretinin, D2-40, and WT-1 and negative staining for CD117, CD34, CD31, PSA, CEA, P40, TTF1, SALL4, and EMA.

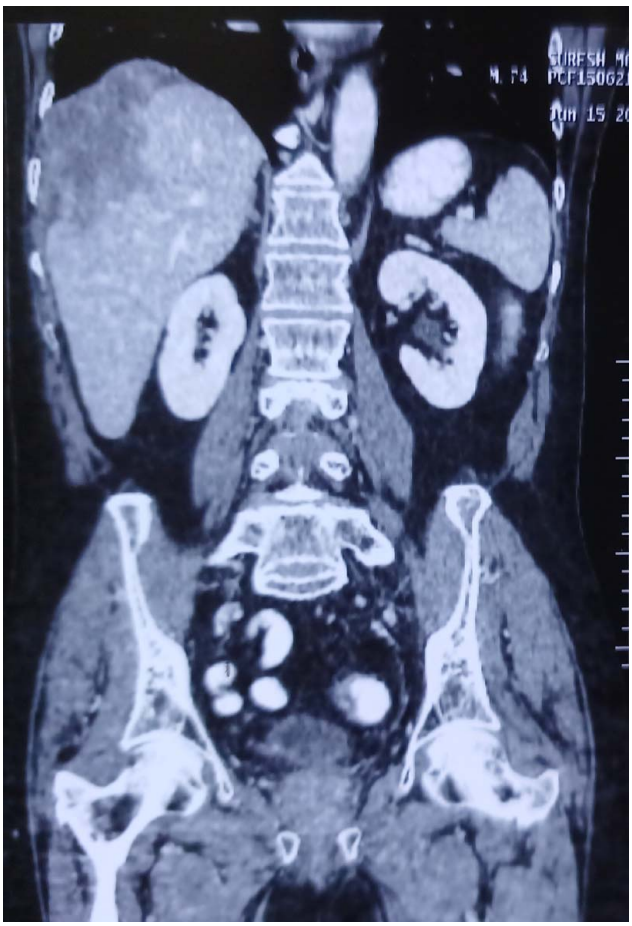

Figure 1. Computer tomography (CT) scan showing 13.6 cm × 11 cm mass in the liver with cystic lesions.

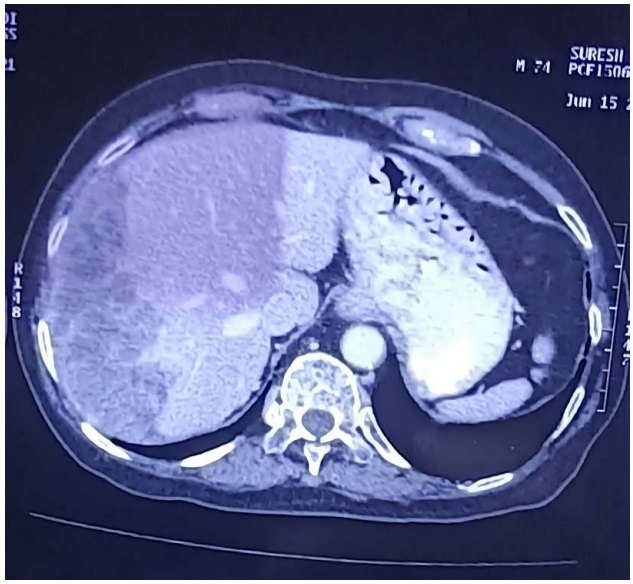

Figure 2. Transverse computed tomography scan. The figure shows a cross-sectional scan of the mass occupying the right lobe of the liver.

The patient was referred to a specialized cancer center for further management. Owing to the advanced stage of the disease and the large size of the tumor, surgical resection was not considered a viable option. The patient was started on palliative chemotherapy with cisplatin and pemetrexed, after reviewing the literature for similar cases and considering the age of the patient, but his condition deteriorated rapidly, and he died within a few months of diagnosis.

Discussion

Sarcomatoid mesothelioma is a rare subtype of mesothelioma that is associated with a particularly poor prognosis. According to the WHO primary intrahepatic neoplasm does not mention

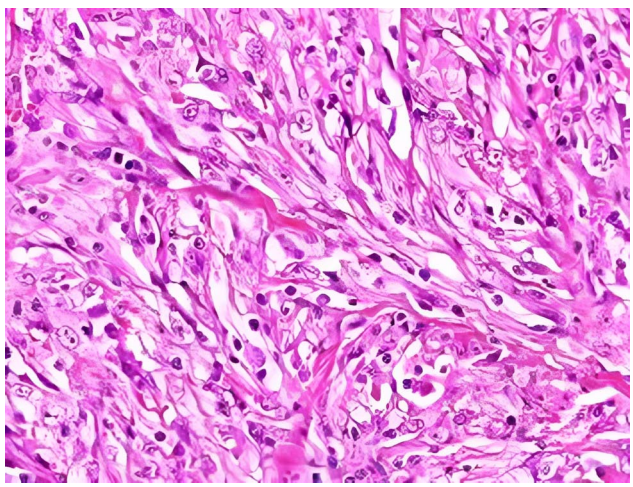

Figure 3. Spindle cells seen on histology feature of a sarcomatoid mesothelioma.

Table 1

Characteristics of patients with PIHMM.

References	Age	Sex	Asbestoses	Size (cm)	Type	Histology (vimenti + calrectin)	Location	Tx.	Relapse	Follow-up
Kottke-Marchant, *et al.*[7]	83	F	N/E	15	Sp.	+	Lt.	Surg	No	3 months
Lenonarda, *et al.*[8]	54	F	Yes	12	Ep.	+	Rt.	Surg	No	2 months
Dong, *et al.*[9]	50	F	No	Multi	Ep.	+	Bilobated	Surg	N/E	N/E
Sakasi, *et al.*[10]	66	M	Yes	4.4	Bp.	+	Rt. (S8)	Surg	No	6 months
Kim, *et al.*[11]	53	M	No	13	Bp.	N/E	Rt.	Surg	DI	N/E
Di Blasi, *et al.*[12]	61	F	N/E	10	Ep.	+	Rt.	Surg	Yes	2 years
Serter, *et al.*[13]	56	F	No	15	Ep.	+	RT (S4, S7, S8)	Surg	No	N/E
Serter,*et al.*[13]	66	M	No	Multi	Bp.	N/E	Bilobated	Surg	No	N/E
Inagaki, *et al.*[14]	68	F	No	7	Ep.	+	Rt. (S7)	BSC	N/E	3 months
Gutgement, *et al.*[15]	62	M	No	5.8	Ep.	+	Rt.	Surg	LNR	5 months
M. Buchholz, *et al.*[16]	62	F	No	5.8	EP.	+	Rt. (S5, S8)	Surg	LNR	20 months
Ghimeeri *et al.*[17]	70	M	Yes	8	Bp	+	Rt (S6)	Surg	No	15 months
Present case	72	M	No	13.6	Sp.	+	Rt. (S5, S6, S7, S8)	BSC	No	N/E

Bp., biphasic; BSC, best supportive care; DI, direct invasion; Ep., endothelial cells; F, female; LNR, *trans*-lymphatic relapse; Lt., left lobe; M, male; N/E, not evaluated; PIHMM, primary intrahepatic mesothelioma; Rt., right lobe; Sp., spindle cells; Surg., surgical; Tx., treatment.

mesothelioma in their list[5]. It is classified into three types: epitheloid, biphasic, and sarcomatoid[6]. The sarcomatoid type is characterized by spindle-shaped cells and a lack of the typical glandular structures seen in other types of mesothelioma. The diagnosis of sarcomatoid mesothelioma is often challenging because it can be difficult to distinguish it from other types of spindle cell tumors.

A review of the 12 cases of mesothelioma is presented in Table 1. This consisted of five male and seven female patients, and only three had a history of exposure to asbestos (25%). All of these had localized tumors at initial diagnosis and with treated by tumor excision but 10 of them relapsed after surgery making it a poor choice of therapy. Most of these cases had the sizes of the lesions varying across cases, ranging from 4.4 cm (Sakasi *et al.*) to 15 cm (Serter *et al.*).Tumor mass as seen on a CT scan. As this is not the case of pleural mesothelioma the chronic inflammation theory, which suggests that inflammation and scarring are the reason for the development in nonasbestos exposure patients would be disapproved. However, the theory of a germline mutation still exists, though we believe that it is spontaneous/idiopathic in this case after a thorough examination and negative findings in the history.

Asbestosis was mentioned in three cases (Lenonarda *et al.*, Sakasi *et al.*, and Ghimeeri *et al.*) and not in six cases (Dong *et al.*, Kim *et al.*, Serter *et al.* (56F), Serter *et al.* (66M), Inagaki *et al.*, and the present case). For two cases (Kottke-Marchant *et al.* and Di Blasi *et al.*), the status was not explicitly mentioned (N/E).

Surgery was the most common treatment option, with nine cases undergoing surgical intervention. The follow-up periods varied, with some cases having no relapse (e.g. Kottke-Marchant *et al.*, Lenonarda *et al.*, Serter *et al.* (56F), and Ghimeeri *et al.*), while others experienced relapse (e.g. Di Blasi *et al.* and Gutgement *et al.*). With this, we can see that for mesothelioma the surgery with or without adjuvant chemotherapy or radiotherapy is to improve the symptoms and survival. However, of these, seven cases were of the epitheliod type (63%) and three were of the biphasic type (27%), and only one case of the sarcomatoid type was seen, which is the same type we present here is. One consistent finding was that in all cases, the tumors were positive for vimentin and calrectin indicating the presence of these markers in the lesions. Which may be used for determining the tumor presence and growth monitoring and further research is required on this topic.

Trimodality therapy is a potentially curative treatment option with neoadjuvant or adjuvant chemotherapy and adjuvant radiotherapy[18]. A systematic review conducted by Cao *et al.* compared outcomes of trimodality therapy studies with the Mesothelioma and Radical Surgery (MARS) trials. Treatment and therapy options are limited to improving quality of life and prolonging survival time. There has been no definitive cure found for mesothelioma.[19] Molecular targeted therapy and immunotherapy may be a potential answer which needs to be evaluated.

Given the rarity of sarcomatoid mesothelioma and the lack of known risk factors, this case presents a diagnostic challenge. Further testing, including molecular profiling of the tumor, was performed to confirm the diagnosis and to guide treatment decisions. In this case, the patient was deemed unsuitable for surgery because of the advanced stage of the cancer and the location of the tumor in the liver. Treatment with chemotherapy and immunotherapy was initiated, but the patient's condition worsened, and he eventually succumbed to the disease.

Conclusion

Despite advances in the treatment of sarcomatoid mesothelioma, much remains to be learned about this disease. One area of active research is the identification of molecular biomarkers that can predict, which patients are most likely to respond to specific treatments. Another area of interest is the development of new therapies, such as targeted drugs and immunotherapies, which may be more effective than traditional chemotherapies. In India, mesothelioma of the liver is a rare disease. The incidence of mesothelioma is relatively low in India compared to Western countries, but it is thought to be under diagnosed due to limited awareness.

Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

K.M., M.J., S.M.: topic conception, data gathering and analysis, manuscript preparation, review, revisions, and editing; G.A.: manuscript preparation, review, revisions, and editing; H.B. and P.T.: data gathering and analysis, manuscript preparation, revisions, and editing.

Conflicts of interest disclosure

The authors declare that they have no conflicts of interest.

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