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CASE REPORT

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Recurrent malignant peritoneal mesothelioma treated by a second resection: A case report

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Key Clinical Message

Malignant peritoneal mesothelioma, a rare and poor prognosis disease, is seldom treated surgically, especially for recurrence. However, early diagnosis and aggressive treatment of primary and recurrent tumors can achieve long-term patient survival.

Abstract

Malignant peritoneal mesothelioma (MPM) is a rare and aggressive tumor, and rarely indicated for surgery, especially for recurrence. In the present case, we report a rare case who could survive long-term after two surgeries in 4 years for MPM.

KEYWORDS

localized mesothelioma, malignant peritoneal mesothelioma, recurrence

1 | INTRODUCTION

Malignant peritoneal mesothelioma (MPM), a rare but lethal malignancy, has a very poor prognosis. The median survival ranges from 5 to 12 months, mainly because making an early preoperative diagnosis is difficult and treatment relatively ineffective.[1] We here present an extremely rare case of a 59-year-old woman who survived long-term after two surgeries in 4 years for MPM. This case report highlights the importance of postoperative follow-up of patients with MPM to ensure early diagnosis of recurrence and enable aggressive treatment.

2 | CASE REPORT

A 59-year-old Japanese woman with a history of hepatic left lateral segmentectomy for a MPM in contact with the hepatoduodenal ligament and partial diaphragmatic resections for three other MPMs on the diaphragm 3 years and 11 months prior to the current presentation[2] was referred to our hospital because an abdominal tumor was found by computed tomography (CT) during postoperative follow-up. The patient had been followed up without chemotherapy after her previous surgery. She had no apparent source of exposure to asbestos at her work or in her residence. On admission, her general condition was unremarkable. Physical examination showed no abnormal findings other than surgical scars. All laboratory data were within normal ranges, including serum concentrations of the tumor markers carcinoembryonic antigen, cancer antigen 19-9, cancer antigen 125, and alpha-fetoprotein protein. Enhanced abdominal CT showed a round hypervascular tumor near the stump of the left hepatic vein (Figure 1). No pleural fluid, ascites, or other nodular lesions were detected in the chest or abdomen. Similar to the CT findings,

abdominal ultrasonography showed a smooth, round, 15-mm tumor that was compressing the middle hepatic vein and inferior vena cava (IVC; Figure 2). Magnetic resonance imaging showed a tumor of lower intensity than the liver on T1-weighted images, and higher intensity than the liver on T2- and diffusion-weighted images (Figure 3A–C). Moreover, positron-emission tomography (PET)-CT revealed that the abdominal tumor exhibited increased uptake of fluorodeoxyglucose metabolism with a maximum of standardized uptake value of 7.4 (Figure 4). Because the lesion was far from the position of the original tumor, we diagnosed metachronous recurrence of MPM preoperatively and resected it. Laparotomy revealed that the tumor was located cranial to the left caudate lobe; however, there was no evidence of invasion of the middle hepatic vein or IVC. We performed tumorectomy with left caudate lobe resection (Figure 5A–C). Hematoxylin–eosin staining showed marked anisokaryosis and structural atypia and immunohistochemistry showed positive staining for CD34, CD31, D-2-40, and calretinin (Figure 6A–H). The pathological findings being similar to those of the original tumor, the final diagnosis was metachronous recurrence of epithelioid MPM. The patient was discharged 10 days after surgery, having had no complications. No adjuvant therapy was administered. There has been no CT and PET-CT evidence of tumor recurrence in the subsequent 16 months.

3 | DISCUSSION

Malignant mesothelioma accounts for only approximately 0.2% of all malignant tumors.[3] MPM is even rarer because malignant mesotheliomas usually arise from the pleura.[4] Surgery is the first-line treatment for MPM. However, the prognosis is characteristically poor with a rapid fatal course. Few patients are candidates for surgery and long-term survival is rarely achieved.[5] We here present a rare case of long-term survival after surgery for recurrence of MPM 4 years after the initial surgery. To the best of our knowledge, no patients who have undergone two surgeries for MPM have previously been reported.

MPM is classified into two types, diffuse or localized. Most patients have the diffuse type with multiple peritoneal nodules and ascites. In contrast, localized MPM is much rarer, usually presenting as a solitary nodular tumor with no ascites and nonspecific symptoms.[2,6] MPM in general is difficult to diagnose early because of its rarity and localized MPM is even more difficult to diagnose. Further complicating the diagnosis of MPM, despite asbestos exposure being the main cause of malignant mesothelioma, patients with MPM are less likely than those with pleural mesothelioma to have a clear history of asbestos exposure.[7] According to one report, only 25% of patients with MPM had a history of asbestos exposure.[8]

Localized MPM has no specific clinical symptoms, tumor markers, or imaging findings and is therefore difficult to diagnose definitively preoperatively. In contrast, the diffuse type of MPM features imaging evidence of ascites and peritonitis.[9] On CT, the tumor is generally

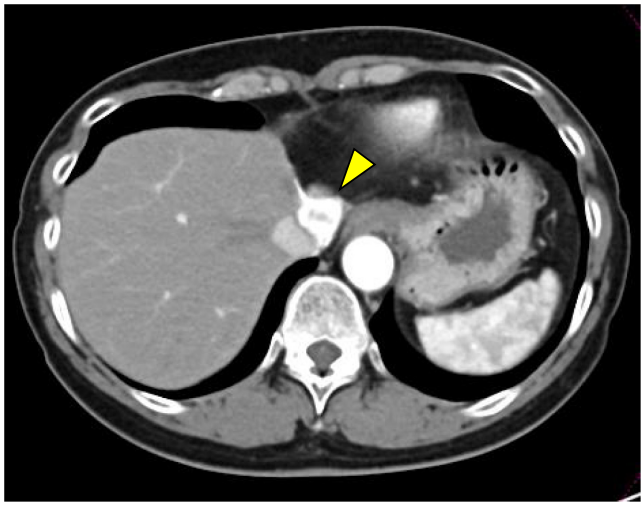

FIGURE 1 Contrast-enhanced abdominal CT image showing a 15-mm diameter hypervascular tumor in contact with the IVC and prior liver transection for left liver lobectomy (yellow arrowhead).

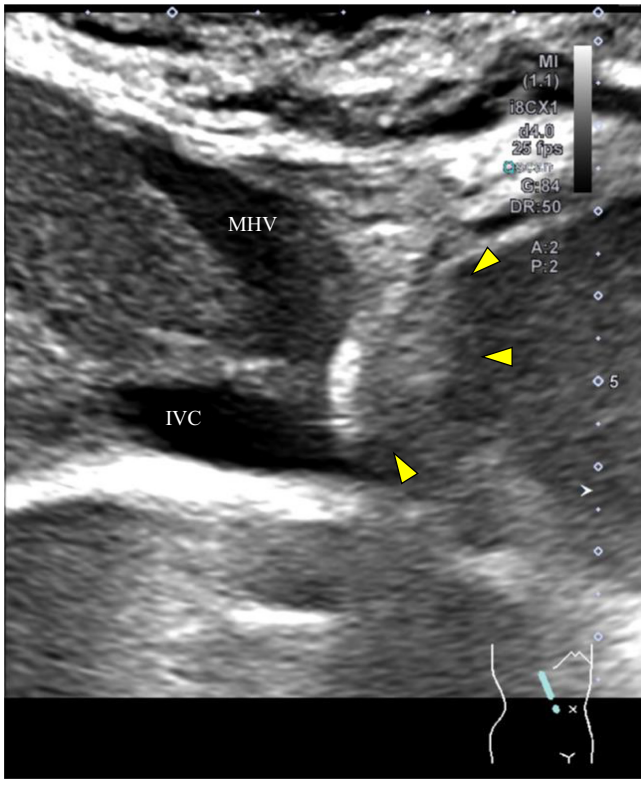

FIGURE 2 Ultrasonography image showing a smooth, round, 15-mm diameter tumor compressing the middle hepatic vein (MHV) and IVC (yellow arrowheads).

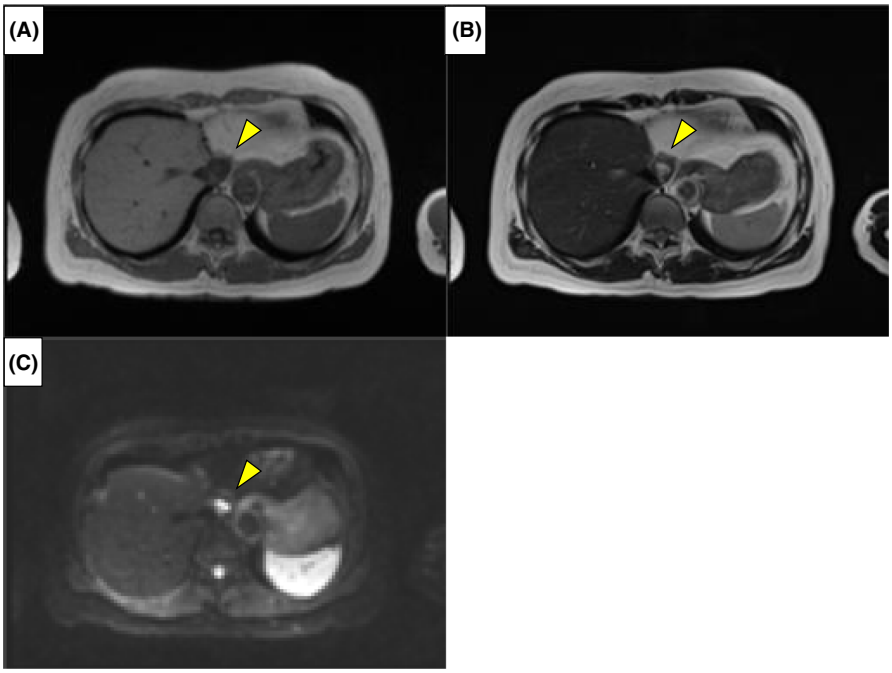

FIGURE 3 Magnetic resonance images showing a tumor with lower intensity than the liver on T1-weighted images (A, yellow arrowhead) and higher intensity than the liver on T2- and diffusion-weighted images (B and C, yellow arrowheads).

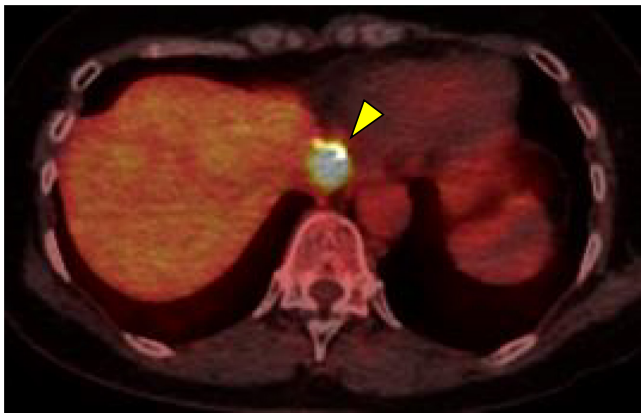

FIGURE 4 Fluorodeoxyglucose-PET image showing a tumor with increased fluorodeoxyglucose uptake (yellow arrowhead).

hypervascular. Furthermore, being a peritoneal tumor, the mass is located on the serosa covering organs. In the case of the liver, it appears as a subcapsular, hypervascular, and hepatic tumor.[2] In addition, because MPM is a peritoneal tumor there may be multiple lesions. Thus, PET-CT examination plays an important role in determining the feasibility of surgical excision.[10] However, there are no imaging findings that are unique to MPM. The possibility of MPM should therefore be considered in addition to commonly encountered diseases. Early treatment can improve the prognosis.

Currently, there is no established standard treatment for MPM. Unlike the diffuse type, for which chemotherapy is generally considered indicated, curative resection is usually the treatment of choice for localized MPM. Because MPM is so rare and localized MPM even rarer, the prognosis after radical resection alone of localized MPM is unknown. Although there are many reported cases of recurrence in the early postoperative period and a subsequent poor prognosis, there are some reports of patients with no recurrence for several years postoperatively.[5,8] Surgery is the treatment of choice, complete resection being expected to cure localized MPM. Despite MPM being so rare that accumulation of cases is difficult, further research, including on the role of chemotherapy, is needed to establish standard treatment for MPM.

This case report highlights the possibility of long-term recurrence-free survival following surgery for recurrence 4 years after initial surgery for MPM. Our patient's recurrence was diagnosed early as a result of 3-monthly CT examinations after the initial surgery, was judged on the basis of PET-CT findings to be operable, and early surgery was performed. When localized MPM recurs, it tends to metastasize like a sarcoma and reportedly has an extremely poor prognosis.[5] In addition, therapeutic strategies for recurrence of MPM are poorly documented. We believe that early diagnosis and treatment, which can be achieved by strict follow-up after surgery, are important in managing patients with MPM.

In conclusion, MPM is very rare, has a poor prognosis, and diagnosis and treatment are often difficult. However, early diagnosis and treatment can lead to long-term survival, especially in patients with localized MPM like our patient. MPM should be included in the differential diagnosis of abdominal tumors. Even if treatment is successful, close follow-up is essential. We are confident that this case report will be useful for those diagnosing and treating this disease.

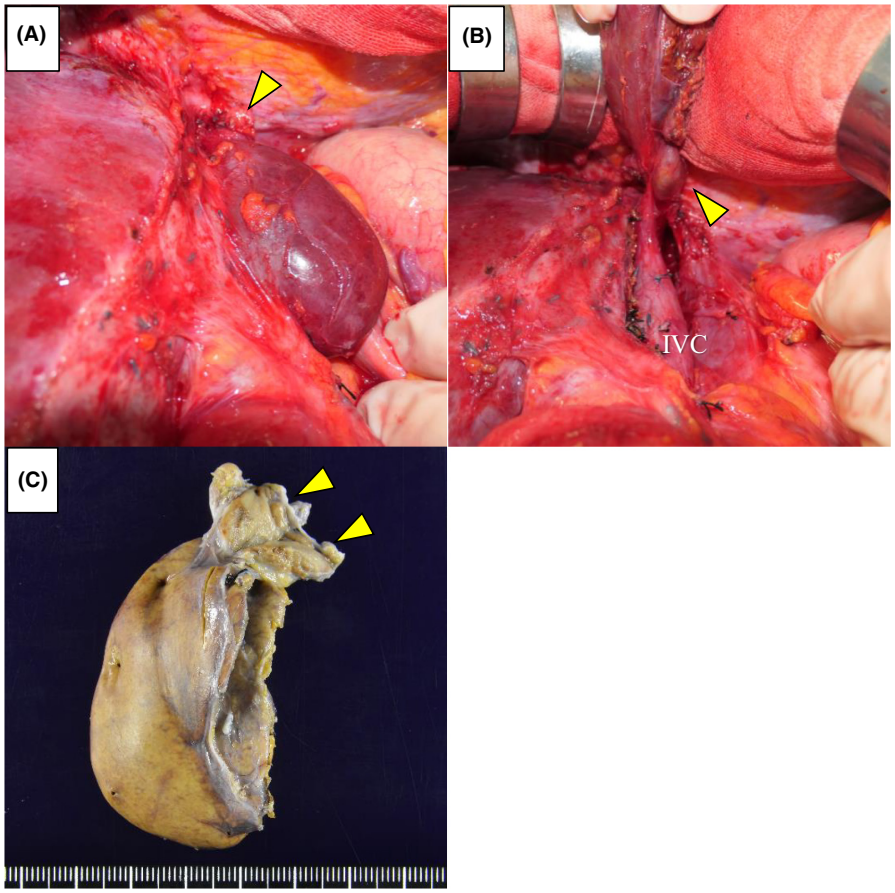

FIGURE 5 Intraoperative photographs and resected specimen showing a tumor located in the top of the left caudate lobe (A, yellow arrowhead), and compression, but not invasion, of the IVC (B, yellow arrowhead). Grossly, the tumor is a well-circumscribed nodular mass with a smooth surface (C, yellow arrowheads).

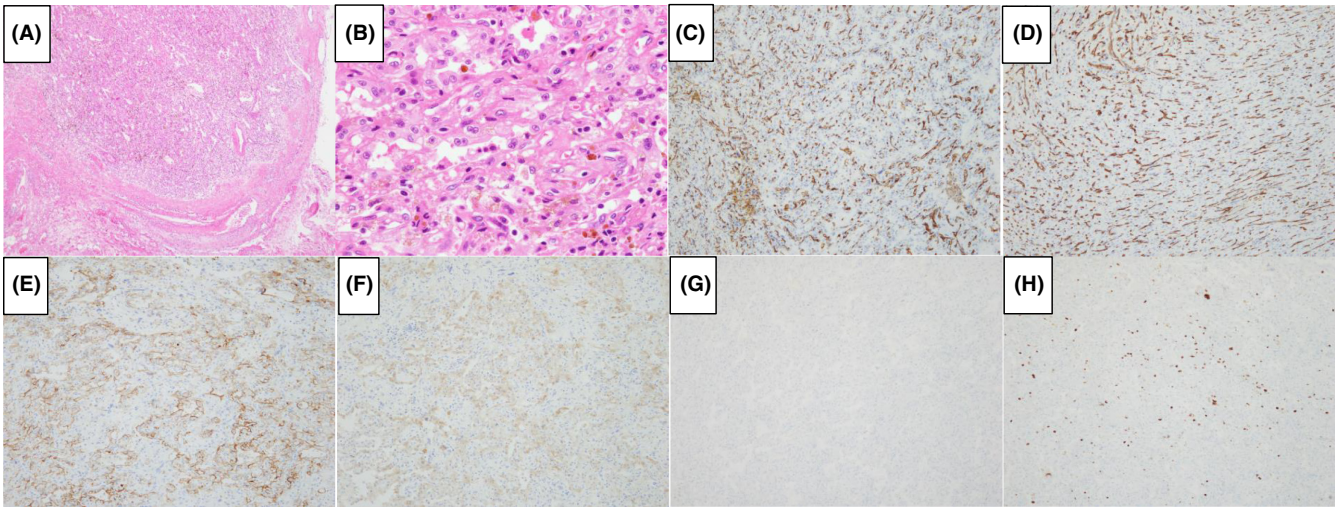

FIGURE 6 Histopathological findings. Photomicrographs showing abundant tumor cells in a papillary form arranged in an epithelial pattern (A, original magnification: ×30; B, original magnification: ×100; all: hematoxylin and eosin stain). The tumor cells stained positively for CD31 (C), CD34 (D), D2-40 (E), and calretinin (F), and negatively for p53 (G). The Ki-67 labeling index was <20% (H) (C–H, original magnification: ×100).

AUTHOR CONTRIBUTIONS

Takashi Miyata: Conceptualization; data curation; visualization; writing – original draft; writing – review and editing. **Hisashi Nishiki:** Conceptualization; writing – review and editing. **Yuki Shinden:** Writing – review and editing. **Shota Motoyama:** Writing – review and editing. **Yuta San-nomiya:** Writing – review and editing. **Hozumi Tamezawa:** Writing – review and editing. **Taigo Nagayama:** Writing – review and editing. **Ryosuke Kin:** Writing – review and editing. **Akifumi Hashimoto:** Writing – review and editing. **Daisuke Kaida:** Writing – review and editing. **Yasuto Tomita:** Writing – review

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CONFLICT OF INTEREST STATEMENT

The authors declare that they have no competing interests. No authors have direct or indirect commercial and financial incentives associated with publication of the article.

DATA AVAILABILITY STATEMENT

Not applicable.

ETHICAL APPROVAL

This case report was approved by the ethics committee of the Kanazawa Medical University.

CONSENT

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

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