BJCR
Brazilian Journal of Case Reports

PCAA
Publicações Científicas de Acesso Aberto

Case Report

Recurrent Multicystic Peritoneal Mesothelioma in a Young Male Patient: A Case Report

Guilherme Bernardes Rosa Eugênio [1,*], Flávio José Müller Filho [1], André Limeira Tenório de Albuquerque [1], Giulia Ferrás [1], Paulo Eduardo Hernandes Antunes [1]

[1] School of Medicine, São Paulo State University (UNESP), São Paulo, Brazil.

* Correspondence: bernardes.med@gmail.com.

Abstract: Multicystic peritoneal mesothelioma (MPM) is a rare benign neoplasm of mesothelial origin, whose pathogenesis is often associated with chronic inflammatory processes, differing etiologically from malignant mesothelioma due to its lack of association with asbestos exposure. We report the case of a young male patient presenting with chronic, nonspecific abdominal symptoms. Imaging evaluation (CT and MRI) revealed multiple septated cystic lesions consistent with the hypothesis of MPM, demonstrating a molding pattern along adjacent peritoneal surfaces (molding sign), without evidence of parenchymal invasion. Despite surgical resection with no macroscopic residual disease (complete excision), the patient developed local recurrence within 24 months. This report emphasizes that, although histologically benign, MPM exhibits a high rate of biological recurrence, requiring a rigorous long-term radiological surveillance protocol and a multidisciplinary therapeutic approach.

Keywords: Benign Cystic Peritoneal Mesothelioma; Peritoneal Neoplasms; Diagnostic Imaging; Local Tumor Recurrence.

Citation: Eugênio GBR, Müler Filho FJ, Albuquerque ALT, Ferrás G, Antunes PEH. Recurrent Multicystic Peritoneal Mesothelioma in a Young Male Patient: A Case Report. Brazilian Journal of Case Reports. 2026 Jan-Dec;06(1):bjcr164.

https://doi.org/10.52600/2163-583X.bjcr.2026.6.1.bjcr164

Received: 26 January 2026
Accepted: 26 February 2026
Published: 3 March 2026

Copyright: This work is licensed under a Creative Commons Attribution 4.0 International License (CC BY 4.0).

1. Introduction

Multicystic peritoneal mesothelioma (MPM), also referred to as multilocular peritoneal inclusion cyst, represents a rare primary neoplastic variant of the peritoneum derived from cystic proliferation of mesothelial cells [1]. Epidemiologically, this condition shows a marked predilection for women of reproductive age; presentation in male patients, as reported in this case, is exceedingly rare and challenges current diagnostic paradigms [1,2]. The global incidence is estimated at 1 to 2 cases per million inhabitants in the general population, accounting for approximately 3% to 5% of all peritoneal mesotheliomas, with no specific sex stratification in most published epidemiological series [2].

Although MPM exhibits predominantly benign biological behavior, its pathogenesis remains controversial. Current theories suggest a reactive-proliferative etiology associated with prior peritoneal insults, such as surgery, endometriosis, or chronic inflammatory processes, distinguishing it from malignant mesothelioma by the absence of a causal link to asbestos exposure [3]. Clinically, the tumor typically presents in an indolent manner with chronic abdominal pain or nonspecific palpable masses.

From a radiological perspective, the differential diagnosis is complex and includes cystic lymphangioma, pseudomyxoma peritonei, and peritoneal pseudocyst. Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) are essential for mapping the

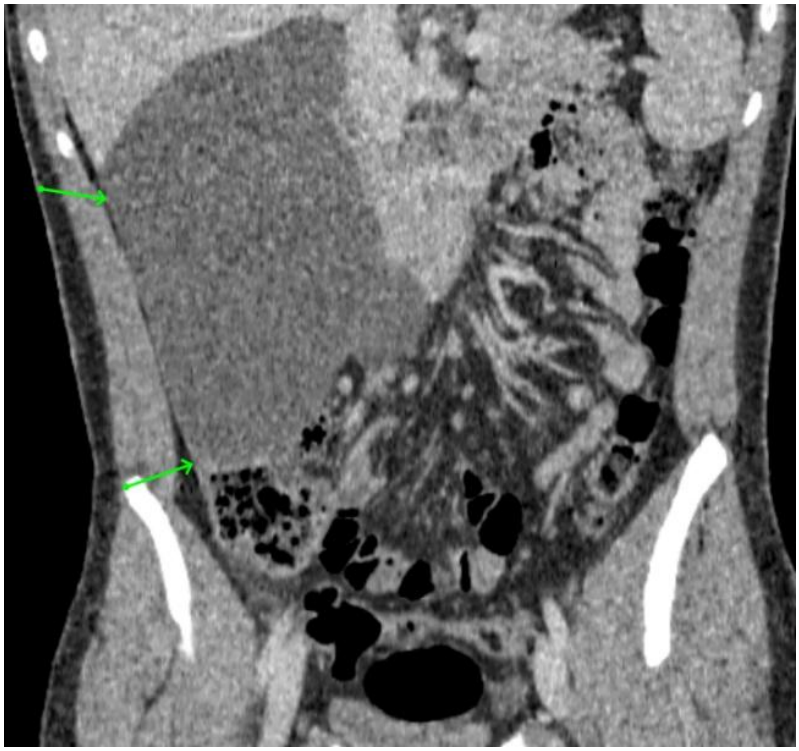
multilocular extent of the lesion and identifying the "molding sign," characterized by filling of peritoneal recesses without infiltration of adjacent solid organs, a finding suggestive of MPM [4]. In particular, the peritoneal topography and molding pattern without infiltrative effect on solid organs favor MPM over lymphangioma, which tends to localize in the mesentery and retroperitoneum and to involve adjacent planes and vascular structures. However, due to the absence of pathognomonic markers, definitive diagnosis remains strictly dependent on histopathological and immunohistochemical analysis. Management is based on complete surgical excision, although the potential for short- and mid-term local recurrence requires a rigorous and prolonged radiological surveillance protocol [2,5].

2. Case Report

A 20-year-old male patient with a history of irritable bowel syndrome presented with chronic, cramping abdominal pain localized to the right hemiabdomen for approximately five years. He had a body mass index (BMI) of 21.9 and no significant laboratory abnormalities. Initially intermittent and of low intensity, the pain progressively became more frequent and severe, associated with abdominal discomfort and distension, particularly after meals. He denied fever, weight loss, or changes in bowel habits. There was no history of abdominal trauma, intra-abdominal infection, or prior surgery.

Initial computed tomography demonstrated a hypodense cystic formation with mild septal enhancement located in the right hemiabdomen, inferior to the right hepatic lobe and extending to the superior cecal region, measuring 16.0 × 12.0 × 12.3 cm, with an estimated volume of 1,228 cm^3 (Figure 1). No solid components, calcifications, or signs of invasion of adjacent solid organs were identified. A molding pattern along adjacent peritoneal surfaces was observed without evidence of parenchymal invasion, consistent with the molding sign.

Figure 1. Coronal reconstruction computed tomography image showing a hypodense lesion (arrowhead), predominantly cystic, located below the right hepatic lobe and extending to the cecal region.

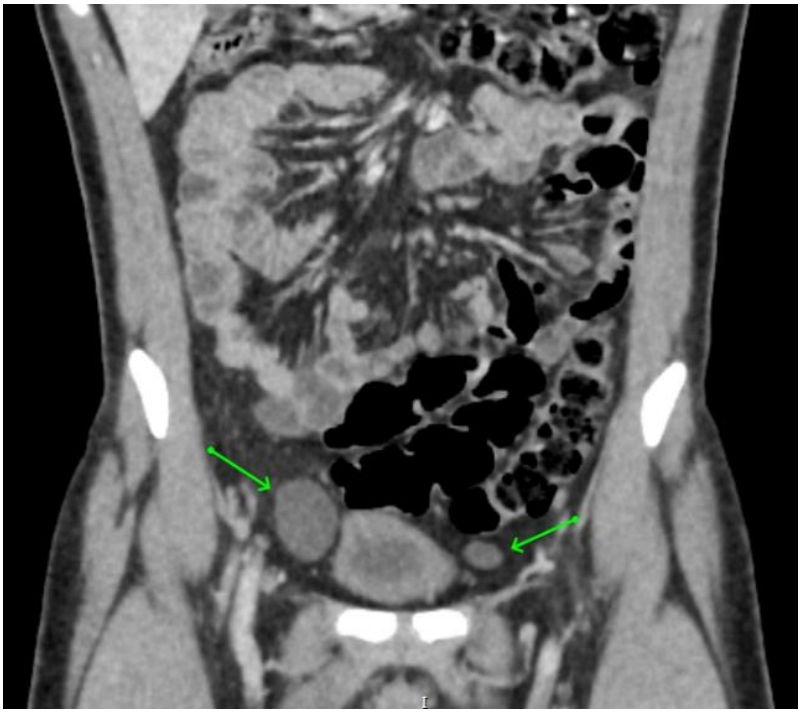


The patient underwent laparotomy with complete excision of the cystic lesion located in the ascending mesocolon. An initial conventional resection was chosen because, in the preoperative setting, the lesion appeared as a seemingly localized cystic mass without tomographic signs of peritoneal dissemination, favoring the hypothesis of a lesion amenable to complete excision. During the surgical procedure, no additional visible peritoneal implants were identified. A systematic inspection of the abdominal cavity was performed, and no macroscopic residual disease was detected. The Peritoneal Carcinomatosis Index (PCI) was not calculated, as there was no preoperative suspicion of disseminated disease.

Histopathological examination confirmed the diagnosis of MPM, with no involvement of the evaluated surgical margins and a preserved lymph node without signs of malignancy. Cytopathological analysis of the lesion revealed mesothelial cells, neutrophils, and red blood cells, without neoplastic cells. Immunohistochemical analysis performed with WT1 and calretinin antibodies showed positive results. In the present case, the available and effectively performed immunohistochemical panel was limited to WT-1 and calretinin, which, in correlation with morphology, supported mesothelial origin. Additional markers for exclusion of differential diagnoses were not performed in this case and are discussed as recommendations in the literature.

After two years of follow-up loss due to the pandemic, the patient returned to resume clinical monitoring. A new contrast-enhanced total abdominal computed tomography scan demonstrated bilateral cystic lesions in the pelvic cavity (Figure 2).

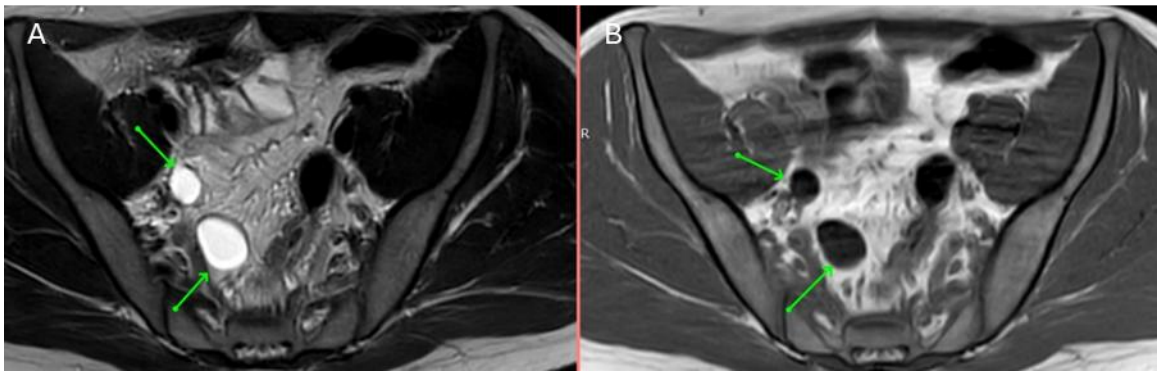
Figure 2. Coronal reconstruction computed tomography image demonstrating two cystic lesions (green arrows) in the pelvic cavity.



Contrast-enhanced pelvic magnetic resonance imaging (MRI) demonstrated two well-defined cystic lesions adjacent to the right iliac vessels, showing hypointense signal on T1-weighted images, hyperintense signal on T2-weighted images, and no significant enhancement after administration of paramagnetic contrast (Figures 3A and 3B). The signal pattern and molding behavior helped exclude cystic lymphangioma in this specific patient. On MRI, the presence of simple fluid content (T1 hypointense and T2 hyperintense signal), the absence of solid components or significant enhancement, and, notably,

the peritoneal distribution with a molding pattern favored MPM. In contrast, cystic lymphangioma typically originates from the mesentery or retroperitoneum and tends to insinuate between planes and vascular structures, a pattern less compatible with the presentation observed in this case.

Figure 3. Magnetic resonance imaging – (A) T2-weighted sequence and (B) T1-weighted sequence demonstrating two well-defined cystic lesions located in the pelvic cavity.



The patient underwent a new surgical procedure for excision of the cysts identified in the pelvis, with complete macroscopic resection. Histopathological examination again confirmed multicystic peritoneal mesothelioma. The second surgery was performed with curative intent at the local level, achieving complete macroscopic resection, although the natural history of the disease carries an inherent risk of further recurrences.

3. Discussion

Multicystic peritoneal mesothelioma (MPM) is a mesothelial neoplasm with an estimated incidence of 1 to 2 cases per million inhabitants. Its occurrence in a young male patient represents a finding of particular academic relevance, given that the disease shows a marked predilection for women of reproductive age [6]. Classically categorized as benign, MPM should be interpreted as a "low-grade" neoplasm due to its local aggressiveness and recurrence rates exceeding 50%, thus requiring a strategic oncologic approach [5]. Although MPM demonstrates predominantly benign biological behavior, its pathogenesis remains controversial. Current theories suggest a reactive-proliferative etiology associated with prior peritoneal insults, such as surgery, endometriosis, or chronic inflammatory processes, distinguishing it from malignant mesothelioma by the absence of a causal link to asbestos exposure [3]. In the present case, no history of abdominal surgery, trauma, intra-abdominal infection, or specific inflammatory process was identified, reinforcing the etiopathogenic heterogeneity described in the literature.

The differentiation among peritoneal cystic masses represents a significant diagnostic challenge due to overlapping findings on computed tomography and magnetic resonance imaging. Cystic lymphangioma is distinguished by its preferential location in the retroperitoneum or mesentery and by the absence of the "molding sign," defined as the ability to fill recesses without parenchymal invasion, a hallmark of multicystic peritoneal mesothelioma. In the present case, both the initial CT and the MRI performed at recurrence demonstrated a peritoneal molding pattern, favoring MPM. In contrast, pseudomyxoma peritonei presents with high-density mucinous ascites and the scalloping sign, characterized by indentations along the serosal surfaces of solid organs such as the liver and spleen [4]. Additionally, peritoneal pseudocyst typically appears as a unilocular lesion lacking epithelial lining and is often associated with a clinical history of prior pancreatitis or recent abdominal surgical interventions.

Definitive diagnosis is strictly based on a comprehensive immunohistochemical (IHC) panel, essential for excluding malignant mimickers and lesions of endothelial

origin. Calretinin is considered the marker of choice, showing strong nuclear and cytoplasmic staining in the cyst-lining cells, while cytokeratin 5/6 (CK 5/6) supports epithelioid mesothelial differentiation [3]. Furthermore, expression of WT-1 (Wilms Tumor 1) assists in distinguishing the neoplasm from purely reactive inflammatory processes.

In the differential diagnosis with malignant mesothelioma, evaluation of BAP1 (BRCA1-associated protein) plays a crucial role: preservation of nuclear expression is characteristic of multicystic peritoneal mesothelioma, whereas loss of expression is typically observed in malignant forms [3,7]. However, BAP1 assessment was not performed in this patient, representing a limitation of the available IHC panel. The diagnosis may also be reinforced by the negativity of exclusion markers: lack of immunoreactivity for CD31, CD34, and D2-40 argues against cystic lymphangioma, while negativity for CEA, MOC-31, and Ber-EP4 excludes ovarian cystadenocarcinomas or peritoneal metastases [3]. These additional markers are described here as recommendations from the literature rather than as findings of the present case.

Clinical management has evolved from simple resection to more aggressive cytoreductive protocols. The recurrence rate after isolated surgical excision is alarmingly high, reaching up to 50% within a mean period of 32 months [5]. The present case corroborates this statistic, with recurrence identified after 24 months. Currently, for multifocal or recurrent cases, the combination of Cytoreductive Surgery (CRS) and Hyperthermic Intraperitoneal Chemotherapy (HIPEC) is considered the preferred strategy. CRS aims at removing all macroscopic disease (peritonectomy), while intraperitoneal infusion of cisplatin or doxorubicin at temperatures of 41–43°C enhances cytotoxicity and drug penetration into serosal surfaces, targeting residual microscopic disease. Studies demonstrate that this approach reduces recurrence rates to below 20%, significantly increasing disease-free survival compared with conventional surgery [5,8]. Despite these data, the applicability of CRS+HIPEC may be limited by case selection, availability of specialized centers, and individualized risk–benefit assessment. In the present case, conventional resection was chosen because the recurrent lesions appeared to have restricted distribution and no macroscopic peritoneal dissemination was observed during surgical exploration. Thus, the adopted approach should be interpreted as treatment with local curative intent, albeit requiring prolonged surveillance and strict follow-up adherence given the risk of additional recurrences.

Although malignant transformation is rare, the chronic nature of the disease and the need for multiple interventions may significantly impact quality of life. Postoperative follow-up should be rigorous, with abdominal and pelvic MRI every six months during the first two years and annually thereafter for at least ten years, considering the possibility of late recurrences [1,2].

4. Conclusion

The present case report reinforces the need for comprehensive diagnostic investigation in young patients presenting with chronic abdominal symptoms, emphasizing that the epidemiological rarity of multicystic peritoneal mesothelioma (MPM) in males should not exclude this diagnostic hypothesis. The patient's clinical course demonstrates the indolent yet recurrent behavior of this pathology.

Recurrence following isolated surgical excision confirms the data reported in the literature and supports the growing evidence of benefits from combined strategies such as cytoreductive surgery (CRS) associated with hyperthermic intraperitoneal chemotherapy (HIPEC) in selected cases, particularly in multifocal or recurrent disease. Rigorous and prolonged radiological follow-up remains essential, especially in view of the risk of silent progression associated with loss to follow-up, thereby ensuring longer disease-free survival and improved quality of life in patients with multicystic peritoneal mesothelioma.

Funding: None.

Research Ethics Committee Approval: We declare that the patient approved participation in the study by signing an informed consent form, and that the study followed the ethical guidelines established by the Declaration of Helsinki.

Acknowledgments: None.

Conflicts of Interest: All other authors declare no conflicts of interest.

References

1. Noiret B, Renaud F, Piessen G, Eveno C. Multicystic peritoneal mesothelioma: a systematic review of the literature. Pleura Peritoneum. 2019 Sep 01;4(3):20190024. doi: 10.1515/pp-2019-0024.
2. Nizri E, Bar-David T, Nissan A, Peretz T, Ben-David M. Multicystic Peritoneal Mesothelioma: Operative and Long-Term Outcomes with Cytoreductive Surgery and Hyperthermic Intra Peritoneal Chemotherapy. Ann Surg Oncol. 2018 Nov;25(11):3724-3730. doi: 10.1245/s10434-018-6712-6.
3. Choudhary S, Fasih N, Mc Innes M, Marginean C. Benign multicystic peritoneal mesothelioma in a male patient. J Radiol Case Rep. 2013 Nov 01;7(11):1-7. doi: 10.3941/jrcr.v7i11.1666.
4. Husain AN, Colby TV, Ordóñez NG, Krausz T, Borczuk A, Cagle PT, Churg A, Cortez FP, Finkelstein S, Fraire AE, Galateau-Salle F, Gibbs AR, Gown AM, Hammar SP, Liz-Guerrero R, Roggli VL, Travis WD, Wick MR. Guidelines for Pathologic Diagnosis of Mesothelioma 2017 Update of the Consensus Statement from the International Mesothelioma Interest Group. Arch Pathol Lab Med. 2018 Jan;142(1):89-108. doi: 10.5858/arpa.2017-0124-RA.
5. Baratti D, Kusamura S, Sironi A, Cabras A, Fumagalli L, Laterza B, Deraco M. Multicystic peritoneal mesothelioma: outcomes and patho-biological features in a multi-institutional series treated by cytoreductive surgery and Hyperthermic Intraperitoneal Chemotherapy (HIPEC). Ann Surg Oncol. 2010 Oct;17(10):2798-2808. doi: 10.1245/s10434-010-1110-x.
6. Pickhardt PJ, Bhalla S. Primary neoplasms of peritoneal and subperitoneal origin: CT findings. Radiographics. 2005 Jul-Aug;25(4):983-995. doi: 10.1148/rg.254045142.
7. Hida T, Hamakawa T, Hayashi T, Igarashi T, Ishida H, Shibuya H, Saitoh S, Kato T, Sakai K, Asano T, Simadzu M, Matsuoka K, Mitsui K, Iwamura M, Hiroshima K. BAP1 immunohistochemistry and p16 FISH to separate benign from malignant mesothelial proliferations. Am J Surg Pathol. 2015 Feb;39(2):201-210. doi: 10.1097/PAS.0000000000000324.
8. Boussios S, Moschetta M, Karathanasi A, Tsiouris AK, Kanellos FS, Tatsi K. Malignant peritoneal mesothelioma: clinical aspects, and therapeutic perspectives. Ann Gastroenterol. 2018 Nov-Dec;31(6):659-669. doi: 10.20524/aog.2018.0305.