Remote Recurrence of Benign Multicystic Peritoneal Mesothelioma

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Abstract

Background: Benign peritoneal cystic mesothelioma (BPCM) is a rare disease entity that arises from mesothelioma cells. We describe a rare case of BPCM recurrence 36 years after its initial presentation.

Case: A 62-year-old woman was referred to an outpatient gynaecologic oncology clinic with an incidental finding of multiple pelvic cysts. She had a preceding history of known BPCM treated with extensive debulking surgery. She presented after 36 years of clinical remission. A repeat laparotomy for a debulking surgical procedure confirmed a recurrence of BPCM.

Conclusion: Our current case represents a woman with a remote recurrence of BPCM after initial optimal debulking surgery. Her clinical presentation of recurrence after 36 years illustrates the need for long-term follow-up and clinical suspicion in symptomatic patients with previously diagnosed BPCM.

Résumé

Contexte : Le mésothéliome kystique bénin du péritoine (MKBP) est une affection rare qui se développe dans les cellules mésothéliales. Nous présentons un rare cas de récurrence survenue 36 ans après la présentation initiale de l'affection.

Cas : Une femme de 62 ans a été dirigée vers une consultation externe en gynéco-oncologie en raison de la découverte fortuite de multiples kystes pelviens. Ses antécédents comprenaient un MKBP traité par une importante chirurgie de réduction tumorale. Ces nouveaux kystes ont été découverts 36 ans après la rémission clinique. Une deuxième chirurgie de réduction tumorale par laparotomie a permis de confirmer la récurrence du MKBP.

Conclusion : Le cas abordé ici est celui d'une femme présentant une récurrence de MKBP longtemps après avoir subi une première chirurgie de réduction tumorale optimale. Cette récurrence, survenue après 36 ans, montre qu'un suivi à long terme et un soupçon clinique de MKBP sont nécessaires chez les patients symptomatiques ayant un antécédent connu de cette affection.

Key Words: Cystic mesothelioma, peritoneal mesothelioma, recurrence

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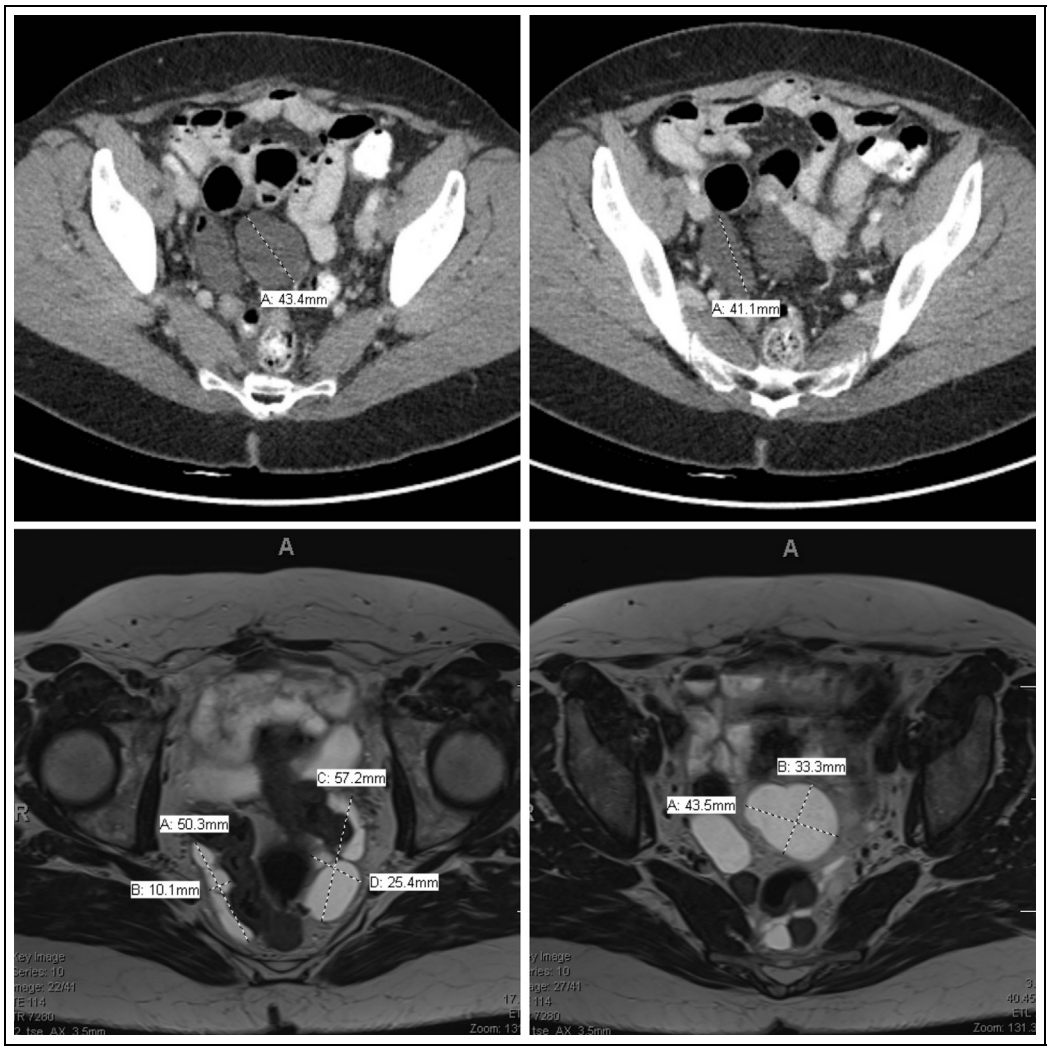
INTRODUCTION

Benign peritoneal cystic mesothelioma (BPCM) is a rare peritoneal tumour arising from the epithelial and mesenchymal components of mesothelial cells. Currently, there are approximately 140 published cases of BPCM, occurring predominantly in women of reproductive age.[1–3] BPCM has been described as the result of a reactive or inflammatory process because of its association with endometriosis, previous abdominal operations, and pelvic inflammatory disease.[1] Currently, the recommended treatment is complete surgical excision with close follow-up, given a local recurrence rate of 41%–50% occurring 2–29 years after initial presentation.[2,4–6] Here, we describe a case report of a patient with a recurrence of BPCM after 36 years of clinical remission.

CASE DESCRIPTION

A 62-year-old woman was referred to an outpatient gynaecologic oncology clinic with an incidental finding of multiple pelvic cysts. After a laparotomy for a benign ovarian cyst at age 17, she underwent two additional laparotomies 9 months apart for BPCM. At age 26, she underwent her first laparotomy for multiple pelvic lesions that were confirmed to be BPCM on the pathology report. However, she experienced a sudden acute onset of lower pelvic pain 9 months later related to the development of a large pelvic peritoneal cyst. Repeat laparotomy, total abdominal hysterectomy, and bilateral salpingo-ophorectomy were completed with the goal of optimal debulking of BPCM. The cysts were found on the pelvic peritoneal surfaces, sigmoid colon, omentum, and

Figure 1. Multiple cystic pelvic lesions on CT (top) and MRI (bottom) scans.



diaphragmatic surface. Again, the same diagnosis was confirmed on the pathology report.

Postoperatively, she had been prescribed hormone replacement therapy. She maintained a state of clinical remission for 36 years. Shortly after discontinuing her hormone replacement therapy, radiologic recurrence was suspected because of an incidental finding of multiple pelvic cysts during investigations for cholestatic symptoms. CT and MRI scans revealed multiple cystic pelvic lesions along the left common iliac vein, right internal iliac artery, right adnexa, pelvic midline, rectosigmoid junction, rectum, and urinary bladder (Figure 1). The largest cyst measured 4.4 × 3.3 cm. The lesions were stable in size over the period of a year. Although the suspicion of malignant transformation of BPCM was low, a debulking operation by laparotomy was offered to confirm our working diagnosis of mesothelioma recurrence. Significant adhesions were encountered during the laparotomy as a result of her previous surgical procedures. The findings described on her CT scan and MRI were confirmed, and all visible disease was excised (Figure 2). Her final pathology report confirmed the diagnosis of benign mesothelial cysts (Figure 3). Histologic examination revealed benign nuclei and a lack of papillary architecture.

DISCUSSION

As described in the literature, BPCM manifested in our patient with extensive involvement of the peritoneal surfaces including the surfaces of the pelvic and abdominal viscera (bladder and bowel) and the retroperitoneum.