Case Report

Multicystic peritoneal inclusion cyst presenting as an incidental abdominal swelling in a middle-aged woman: A rare case report

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ABSTRACT

Multicystic peritoneal inclusion cyst (PIC), historically designated benign cystic mesothelioma, is an exceptionally rare, benign proliferation of peritoneal mesothelial cells. We report a middle-aged woman in whom a multilocular cystic peritoneal swelling was detected as an incidental finding on abdominal ultrasonography and computed tomography. Serum tumor markers (Cancer Antigen 125, Carcinoembryonic Antigen, and Carbohydrate Antigen 19-9) were within normal limits, and both ovaries and the uterus were visualized separately and appeared normal on imaging. The lesion lacked solid components, septations with vascularity, or any features to suggest malignancy on pre-operative imaging. Diagnostic laparoscopy, converted to open surgery on account of the extent of peritoneal involvement, allowed complete surgical excision. No spillage of cyst contents occurred, and no residual lesion was identified on thorough peritoneal inspection. Histopathological examination confirmed the diagnosis of PIC, demonstrating multiple cystically dilated spaces lined by a single layer of bland mesothelial cells with characteristic hobnail morphology, without atypia, mitotic activity, or stromal invasion. This case illustrates the diagnostic challenge posed by peritoneal cystic lesions, the indispensable role of histopathology in their definitive characterization, and the importance of complete surgical excision given the recognized propensity for local recurrence.

Key words: Hobnail mesothelial cells, Peritoneal inclusion cyst, Abdominal swelling, Benign cystic mesothelioma

Peritoneal inclusion cyst (PIC), also known historically as benign cystic mesothelioma or multicystic peritoneal mesothelioma, is a rare, benign mesothelial proliferation arising within the peritoneal cavity [1]. Since its original description by Smith and Mennenmeyer in 1979, fewer than 200 cases have been recorded in the published literature, making this one of the least frequently encountered lesions in surgical pathology practice [2,3]. The condition predominantly affects women of reproductive and perimenopausal age, though cases have been described across a broad age range and, rarely, in men [4]. Recognized predisposing factors include a history of abdominal or pelvic surgery, endometriosis, pelvic inflammatory disease, and other conditions associated with chronic peritoneal irritation; however, a significant proportion of patients lack any identifiable predisposing factor [1,5]. The pathogenesis remains a subject of debate; the reactive hypothesis regards PIC as an exuberant mesothelial response to peritoneal injury, whereas the neoplastic hypothesis interprets it as a true low-grade neoplasm of mesothelial origin [3,5].

Access this article online	
Received - 17 June 2026 Initial Review - 08 July 2026 Accepted - 28 July 2026	Quick Response code
DOI: 10.32677/ijcr.v12i8.8345	

The rationale for presenting this case is threefold. First, the incidental discovery of PIC in a middle-aged woman, without any classical predisposing history of endometriosis or prior pelvic surgery, underscores the breadth of clinical scenarios in which this diagnosis must be considered. Second, the multilocular cystic morphology visible on pre-operative imaging closely resembles several sinister conditions, including lymphangioma, ovarian serous cystadenoma, pseudomyxoma peritonei, and even cystic malignancy, creating a genuine diagnostic dilemma that can only be resolved by histopathological examination. Third, the surgical decision, a laparoscopy converted to open excision, and the pathological findings of this case provide a practical illustration of the diagnostic and therapeutic principles applicable to peritoneal cystic lesions. Awareness of this entity among clinicians, radiologists, and pathologists is essential to avoid unnecessary radical surgery on the one hand and under-treatment with subsequent recurrence on the other hand.

CASE REPORT

A 49-year-old woman was referred for evaluation of an incidentally detected abdominal cystic swelling.

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The lesion had been identified on ultrasonography performed during a routine health check-up; the patient herself had been asymptomatic and denied abdominal pain, distension, nausea, vomiting, altered bowel habits, or urinary symptoms. There was no history of prior abdominal or pelvic surgery, endometriosis, or pelvic inflammatory disease. Menstrual history was unremarkable.

On general examination, the patient was in good health, with a body mass index of 22 kg/m^2 and no known comorbidities (no diabetes mellitus, hypertension, or other chronic illness). Vital signs were within normal limits. Abdominal examination revealed a soft, non-tender abdomen with no palpable organomegaly. A vague, non-tender fullness was appreciated in the lower abdomen on deep palpation, without guarding or rigidity.

Abdominal and pelvic ultrasonography demonstrated a multilocular cystic mass in the lower abdomen and pelvis, containing clear anechoic fluid within thin-walled locules. No internal echoes, solid nodules, or internal vascularity on Doppler interrogation were identified. The uterus and bilateral ovaries were visualized separately and appeared normal. Contrast-enhanced computed tomography (CT) of the abdomen and pelvis confirmed a large, well-defined, multiloculated cystic lesion occupying the lower abdomen and pelvis, with imperceptibly thin walls and no enhancing solid component, mural nodule, calcification, or associated lymphadenopathy. The lesion appeared to arise from the peritoneum rather than from either ovary. CT attenuation values of the cyst contents were consistent with simple fluid. Serum tumor markers, including Cancer Antigen 125, Carcinoembryonic Antigen, and Carbohydrate Antigen 19-9, were within normal limits.

The patient was planned for diagnostic laparoscopy. Intraoperatively, multiple translucent, thin-walled cysts were identified arising from the parietal and visceral peritoneum, clustered predominantly in the pelvis and lower abdomen, with involvement of the adjacent omental and mesenteric peritoneal surfaces. Both ovaries and the uterus were visualized and appeared entirely normal. Given the diffuse peritoneal extent of the lesion and the technical difficulty of achieving complete laparoscopic excision, the procedure was converted to open surgery. Complete excision of all visible cystic components was performed with no spillage of cyst contents inside her body. The peritoneal cavity was inspected thoroughly, and no residual lesion was left behind.

The excised specimen comprised multiple translucent, thin-walled cysts of varying sizes (ranging from 0.5 cm to several centimeters in maximum diameter), containing clear, watery fluid. The tissue was received in fragments within the histopathology section, as the surgeon removed it intact; however, he visualized himself while cutting open in the operating theater's side room. The cyst walls were smooth on both inner and outer surfaces, without papillary excrescences, intra-cystic solid areas, hemorrhage, or necrosis. The walls were soft, pliable, and non-calcified. Representative sections were submitted for histopathological processing.

Microscopic examination of hematoxylin and eosin-stained sections demonstrated multiple cystically dilated spaces separated by fibrocollagenous stroma. The cysts were lined by a single layer of elongated to cuboidal mesothelial cells exhibiting a distinctive hobnail appearance, characterized by apical cytoplasmic bulging of the lining cells into the cyst lumen. The nuclei were bland, round to oval, and uniform, without pleomorphism, hyperchromasia, or prominent nucleoli. No epithelial stratification, mitotic figures, papillary formations, or psammoma bodies were identified. Crucially, there was no evidence of stromal invasion. The intervening stroma was fibrocollagenous and devoid of granulomas, significant acute or chronic inflammation, or necrosis. No evidence of malignancy was identified in any of the sections examined (Fig. 1). Immunohistochemistry (IHC) was not performed due to cost restraints. The diagnosis was based on these characteristic morphological features, which are widely regarded as diagnostic of PIC when classical hobnail mesothelial lining and absence of atypia are present. A final histopathological diagnosis of multicystic PIC was made after clinic-radiological correlation.

DISCUSSION

PIC is a rare and diagnostically challenging entity whose precise biological nature has long been debated. The central question, whether PIC represents an exuberant reactive proliferation of mesothelial cells in response to peritoneal injury, or a true low-grade neoplasm of mesothelial origin, remains unresolved [3,5]. This uncertainty is reflected in the evolving nomenclature: The World Health Organization Classification of Tumours (5th edition, 2020) introduced the broader umbrella term "peritoneal inclusion cyst" to encompass lesions at the reactive end of the spectrum, while retaining "mesothelioma" for cases in which cytological atypia is demonstrable [5]. The two designations are therefore not mutually exclusive but reflect position along a biological

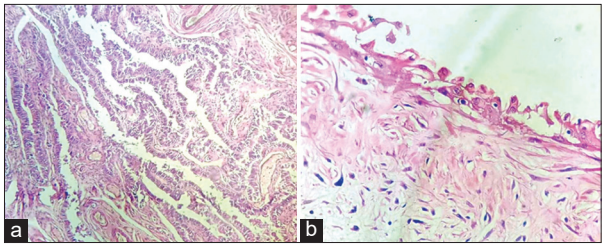


Figure 1: (a) Photomicrograph showing cystically dilated spaces of varying sizes separated by fibrocollagenous stroma, demonstrating the characteristic multilocular architecture of a peritoneal inclusion cyst (haematoxylin and eosin [H&E], ×40); (b) Photomicrograph showing cyst wall lined by a single layer of cuboidal mesothelial cells showing hobnail morphology with apical cytoplasmic protrusion. Nuclei are bland and uniform. No stromal invasion identified (H&E, ×200)

continuum. In the present case, the entirely bland histology, single-layered mesothelial lining, no atypia, no mitoses, no stromal invasion, places the lesion firmly at the benign, reactive pole of this spectrum, consistent with the designation PIC.

The clinical profile of PIC, incidental discovery in an otherwise well, middle-aged woman with no prior pelvic surgery or endometriosis, is atypical for the classical presentation but has been reported [1,4]. Most of the cases present with vague lower abdominal pain or pelvic pressure attributable to the mass effect of accumulating cysts; a purely incidental presentation, as in this patient, reflects the indolent natural history of many lesions and the increased detection of abdominal pathology through widespread use of cross-sectional imaging [2,4].

On imaging, PIC typically appears as a multiloculated cystic pelvic or abdominal mass with thin, non-enhancing walls, no internal solid component, and no associated lymphadenopathy, findings that were reproduced faithfully in the present case [2,6]. Nevertheless, the imaging differential remains broad and includes lesions of fundamentally different biological behavior, making histopathological confirmation essential before definitive management is planned.

Histopathologically, the hallmark of PIC is the presence of multiple cystically dilated spaces lined by a single layer of bland mesothelial cells exhibiting the characteristic hobnail morphology, against a background of fibrocollagenous stroma and without evidence of invasion or atypia [1,3]. The hobnail appearance created by apical cytoplasmic protrusion of the lining cells, represents a pattern of reactive mesothelial change rather than a feature of neoplastic transformation [1,7]. These features, combined with the absence of a genuine epithelial lining (ruling out lymphangioma and serous cystadenoma), the absence of laminated membranes (ruling out hydatid cyst), and the absence of mucin (ruling out pseudocyst and pseudomyxoma peritonei), were decisive in establishing the diagnosis in the present case. Differential diagnosis of a multilocular peritoneal cystic lesion is shown in Table 1.

Several recently published case reports illustrate the diverse clinical contexts in which PIC may present. Killoran *et al*. described a PIC in a young male managed with debulking surgery, highlighting that this entity is not confined to women of reproductive age [3]. Wassef *et al*. reported a case in which the mass was densely adherent to adjacent viscera, requiring meticulous dissection to achieve complete resection [8]. Al-Saghir *et al*. described a large multilocular PIC that was initially misdiagnosed as a lymphangioma on imaging, underscoring the reliance on histopathology for definitive diagnosis [9]. Patel *et al*. reported an incidental PIC discovered concurrently with mucinous colon adenocarcinoma, illustrating the potential for diagnostic confusion with peritoneal carcinomatosis in patients with a known visceral malignancy [10]. These reports, together with the present case, reinforce the wide demographic and clinical spectrum of this rare entity and the consistent, central role of histopathology, with or without IHC, in establishing the diagnosis.

The operative findings in this case, diffuse peritoneal cystic involvement necessitating conversion from laparoscopy to open surgery, are consistent with published series documenting widespread peritoneal distribution in a proportion of PIC cases [2,6]. Complete surgical excision remains the recommended first-line treatment, as it both confirms the diagnosis histologically and removes the bulk of the disease [1,2]. Recurrence rates of up to 50% have been reported, particularly following incomplete excision or in cases with extensive peritoneal involvement [3,7]. In the present case, complete macroscopic clearance was achieved, and the patient was counseled regarding the need for long-term clinical and radiological surveillance. Malignant transformation to epithelioid mesothelioma is documented but remains exceedingly rare [3,7].

Table 1: Differential diagnosis of a multilocular peritoneal cystic lesion

Diagnosis	Key imaging features	Histology	Distinguishing feature	Excluded in this case
PIC	Thin-walled multilocular cysts; no solid component	Single layer bland mesothelial cells; hobnail morphology; no invasion	Mesothelial lining; fibrocollagenous stroma	— (confirmed diagnosis)
Lymphangioma	Multilocular cystic; may show chyle density	Endothelial lining (CD31/ D2-40 positive); lymphoid aggregates in wall	Endothelial not mesothelial lining	No endothelial lining or lymphoid aggregates identified
Serous cystadenoma (ovarian origin)	Cystic adnexal mass; may have papillae	Columnar ciliated epithelium; papillary projections possible	Ovarian origin; tubal-type epithelium	Both ovaries normal; no papillae or columnar epithelium
Pseudocyst/ mesenteric cyst	Unilocular or multilocular; no internal vascularity	No epithelial lining (fibrous wall only)	Absence of any lining layer	Definite mesothelial lining present
Hydatid cyst	Cyst-within-cyst; rim calcification	Laminated eosinophilic membrane; scolices/hooklets	Parasitic elements	No laminated membrane or parasitic elements
Pseudomyxoma peritonei	Mucinous ascites; scalloping of liver/ spleen	Mucin pools; mucinous epithelium; appendiceal primary usual	Mucin production	No mucin; no appendiceal lesion

PIC: Peritoneal inclusion cyst

Several adjuncts to surgery have been described in the management of recurrent or unresectable PIC, including hormonal therapy (progesterone, tamoxifen), interferon-alpha, and, in selected cases, cytoreductive surgery combined with hyperthermic intraperitoneal chemotherapy [6,7]. None of these modalities has been evaluated in prospective trials, and treatment decisions for recurrent disease must be individualized.

This report has certain limitations. IHC was not performed to further confirm mesothelial origin, primarily owing to cost constraints; the diagnosis instead relied on the characteristic hobnail morphology and bland cytology of the lining cells. Detailed long-term post-operative follow-up data are not yet available at the time of this report, given the recognized recurrence rate of up to 50% for this entity; the patient remains under planned clinical and radiological surveillance, and follow-up findings will be reported as they become available.

CONCLUSION

PIC is a rare but clinically important peritoneal lesion whose multilocular cystic morphology can convincingly mimic a range of conditions from the innocuous to the malignant; its definitive diagnosis rests entirely on histopathological confirmation, and complete surgical excision, guided by a high index of clinical suspicion, remains the cornerstone of management, with long-term surveillance warranted given the propensity for local recurrence.

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Funding: Nil; Conflicts of interest: Nil.

How to cite this article: Warpe BM, Joshi SS, Chavan JR. Multicystic peritoneal inclusion cyst presenting as an incidental abdominal swelling in a middle-aged woman: A rare case report. Indian J Case Reports. 2026;12(8):569-572.