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Multilocular Peritoneal Inclusion Cysts Presenting in the Setting of Ischemic Colitis and Pneumoperitoneum

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Background	A 43-year-old non-verbal patient with cerebral palsy and multilocular peritoneal inclusion cysts presented as pneumoperitoneum on CT imaging. We discuss this case and a review of the pathology, clinical presentation, and management of multilocular peritoneal inclusion cysts.
Summary	A 43-year-old non-verbal patient with cerebral palsy presented with an increase in the frequency of bowel movements and hematochezia, as reported by her guardian. CT imaging revealed a pneumoperitoneum with portal vein gas suspicious for ischemic colitis with viscus perforation. The patient underwent exploratory laparotomy revealing multiple cystic structures on the omentum, with the greatest concentration at the hepatic flexure. Final pathology showed multilocular peritoneal inclusion cysts. Multilocular peritoneal inclusion cysts are a rare disease most commonly affecting reproductive females. These cysts are commonly misdiagnosed or hard to identify on conventional imaging modalities. This case presents multilocular peritoneal inclusion cysts in a 43-year-old non-verbal female with cerebral palsy. This case explores a review of the presentation, histological findings and diagnostic considerations, and the management of multilocular peritoneal inclusion cysts.
Conclusion	Multilocular peritoneal inclusion cysts are a diagnostic challenge as they are rare and more commonly seen in the pelvic region. However, when present in the upper abdomen, they can be difficult to distinguish from pneumoperitoneum on CT imaging, forcing clinicians to initially manage patients with exploration—even though it is a benign disease process.
Key Words	multilocular peritoneal inclusion cysts; benign multicystic peritoneal mesothelioma; pneumoperitoneum

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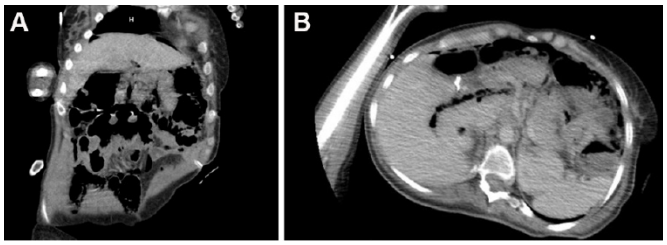
Case Description

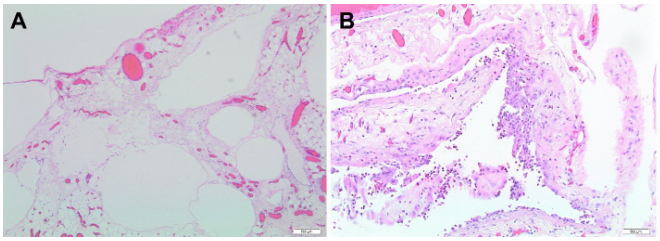
A 43-year-old woman with cerebral palsy presented to the emergency department due to increased bowel movements and hematochezia over a couple of days before presentation. The patient is non-verbal and cannot give an accurate history of physical complaints. Her guardians acknowledge that the patient has been reaching for her stomach and having facial expressions reflecting pain. The patient has a gastric tube and a jejunal feeding tube. The patient had an extensive medical history, including gastroparesis, GERD, ischemia of small intestines, and small bowel obstruction. She also had a history of multiple abdominal surgeries, including laparoscopic cholecystectomy and exploratory laparotomy. At the exam, the patient was afebrile, hemodynamically stable with a minimally elevated WBC of 11,400. A CT scan was completed showing portal venous air extending to the intrahepatic and extrahepatic portal vein and a small amount of intraluminal gas in the distal SMA branch vessels (Figure 1). There were also edematous changes in the small and large bowel mesentery with adjacent free air and free intraperitoneal air under the diaphragm. Based on the CT findings and concern for ischemic colitis with perforation, an exploratory laparotomy was performed.

Figure 1. CT Scan Images Showing A) Pneumoperitoneum with Free Air under Diaphragm and B) Surrounding Colon. Published with Permission



Exploratory laparotomy demonstrated multiple omental cysts diffusely throughout the omentum with the largest collection about the hepatic flexure (Figure 2). Grossly, the cysts appeared to be primarily air-filled with little clear, watery fluid. The serosal surface of the bowel was normal with no signs of ischemia. A partial omentectomy was performed near the hepatic flexure and sent for pathological analysis. Pathologic analysis confirmed the presence of multilocular peritoneal inclusion cysts or benign cystic mesothelioma.

Figure 2. Exploratory Laparotomy Demonstrating Multiple Omental Cysts Diffusely Throughout the Omentum. Published with Permission



A) Low-power view showing multiloculated cystic structures in omentum; B) High-power view showing with single layer of mesothelial cells lining cystic spaces

Postoperative day 2, the patient was evaluated via sigmoidoscopy showing patchy erythema and induration of the distal transverse and descending and proximal sigmoid colon. Biopsy pathology showed fragments of benign colonic mucosa with mild, diffuse chronic inflammation with extravasated red blood cells suggestive of early signs of mild ischemic colitis.

Discussion

Multilocular peritoneal inclusion cysts or benign multicystic peritoneal mesothelioma is a rare diagnosis that typically occurs in the peritoneal cavity in women of reproductive age.[1–4] These cysts can be seen on imaging and identified as multiseptated cystic masses[2]; however, they require pathologic biopsy for definitive diagnosis.[1,3] On gross examination, these cysts tend to have a smooth lining and may be filled with a yellow watery or gelatinous material.[1] Whereas on microscopic sectioning, the cysts are lined by a single layer of benign mesothelial cells.[1] Benign cystic mesothelioma implies histological evidence of nuclear atypia, whereas peritoneal inclusion cyst implies the normal nuclear appearance of the mesothelial cells lining the cyst.[4] Current evidence suggests the pathogenesis is inflammatory. Some cases will contain markers of acute and chronic inflammation, including abundant fibrin, dense bands of granulation tissue with recent and remote hemorrhage along the cyst wall.[1]

These cysts are most commonly found attached to pelvic organs, imitating an ovarian cyst.[1,2] The presentation can vary from asymptomatic to lower abdominal pain, and some have been cited to present as acute abdominal appendicitis.[5,6] Many forms of imaging are available to evaluate multilocular peritoneal inclusions cysts, including sonography, CT, and MRI. However, there is no consensus on

the best diagnostic approach as they are commonly incidental findings and challenging to distinguish from other types of cysts or pseudocysts.[3] Definitive diagnosis depends on a histological specimen obtained via laparoscopy, laparotomy, or image-guided biopsy.[1–3]

There is currently no good evidence supporting the preferred management of multilocular peritoneal inclusion cysts.[3] Multiple treatment modalities have been utilized in other cases, including observation, Ultrasound- or CT-guided aspiration, surgical resection, and even chemotherapy in refractory and symptomatic cases.[3] Current literature suggests that observation is a reasonable option for asymptomatic patients as it is a benign lesion. Treatment options such as surgical excision and aspiration can be considered when patients become symptomatic; however, recurrence can occur.[3] If surgical resection is utilized, there is a tendency for benign multicystic peritoneal mesothelioma to recur in up to 50 percent of cases.[2]

In this case, the patient has a medical history of cerebral palsy, making her non-verbal. This scenario challenges clinicians as the patient cannot provide an adequate history and accurate physical exam. Thus, clinicians must rely heavily on imaging studies for medical decision-making. CT imaging of this patient showed potential signs of free intraperitoneal air and portal vein gas suggesting ischemic colitis with viscus perforation. The patient was taken to the operating room for an exploratory laparotomy due to the concern for perforated viscus; however, no perforation or signs of gross ischemia were found. With a negative exploration for perforation, it is reasonable to conclude that the air seen on CT imaging surrounding the colon may have been due to the presence of the multilocular inclusion cysts. This finding in the area of the hepatic flexure is an uncommon presentation as these cysts are most commonly found around the pelvic organs.[1,2]

Conclusion

This case demonstrates the difficulties clinicians face when relying on imaging and laboratory studies to guide their medical decision-making. Multilocular peritoneal inclusion cysts in the upper abdomen are a diagnostic challenge as it is a relatively rare disease more commonly seen in the pelvic region. However, when present in the upper abdomen, they can be difficult to distinguish from pneumoperitoneum on CT imaging, inclining clinicians to initially manage patients with exploration to rule out viscus perforation, even though the cysts themselves are a benign disease process.

Lessons Learned

Peritoneal inclusion cysts can present similarly to pneumoperitoneum on CT imaging and therefore should be considered in the differential diagnosis of pneumoperitoneum. Patients with and without documented peritoneal inclusion cysts may be difficult to distinguish between these cysts and viscus perforation. Regardless, in this clinical setting, an exploratory laparotomy was required to rule out the presence of viscus perforation.

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