Laparoscopic excision of benign multicystic peritoneal mesothelioma

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Benign multicystic peritoneal mesothelioma is a rare disease that arises from the peritoneal mesothelium. We report on a 52-year-old woman who presented with a large abdominal multicystic mass presumed to be a pancreatic pseudocyst. Laparoscopic exploration revealed a multicystic mass with area of calcification originating from the lesser curvature of the stomach. The whole tumor was successfully excised laparoscopically. Histopathology revealed benign multicystic peritoneal mesothelioma with an area of calcification. Treatment by a minimal access approach allowed the patient to recover rapidly with a short convalescence. Our case confirms the feasibility and safety of a minimal access surgical approach to a rare pathological entity.

Keywords:
abdominal cyst, abdominal mass, laparoscopy, peritoneal mesothelioma

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Introduction

Benign multicystic peritoneal mesothelioma (BMPM) is a rare disease with annual incidence of 0.15/100 000 arising from the peritoneal mesothelium [1,2]. We report, to the best of our knowledge for the first time in the surgical literature, a case of a BMPM arising from the lesser curvature of the stomach successfully resected by laparoscopy.

Case report

A 52-year-old female patient presented to us by a history of recurrent attacks of dull aching pain in the epigastrium of about 2 years duration. This pain was radiating to the back, increased by food ingestion, and was associated with nausea and vomiting. There was no history of previous surgical operations or other comorbidities. Her abdominal examination was irrelevant with no palpable masses. All laboratory results were within normal ranges. Serological markers for hydatid cyst were negative. Carcinoembryonic antigen was 3.2 ng/ml and carbohydrate antigen 19-9 was 2 U/ml. Esophagogastroduodenoscopy was ordered and showed extrinsic compression on the posterior aspect of body of the stomach, but there were no evident mucosal abnormalities. Abdominal computed tomography (CT) revealed a 7 × 5 cm ill-defined cystic lesion in the lesser sac containing areas of calcification. The lesion was related to anterior aspect of the pancreas with no line of separation. CT diagnosis was mostly a pancreatic pseudocyst (Fig. 1).

The patient was submitted for laparoscopic exploration. A nasogastric tube was placed. The patient was placed in the French position with the surgeon standing between legs. Five ports were used (Fig. 2): a 10-mm port above the umbilicus (for camera), another 10-mm port at the left midclavicular line for the surgeon's right hand, and three 5-mm ports (one at the right midclavicular line for the surgeon's left hand, one at the left anterior axillary line for the assistant, and one just below the xiphisternum for liver retraction).

Laparoscopic examination revealed a grape-like cystic lesion located beneath, but not attached to, the left liver lobe. The lesion was covered by a thin layer of the gastrohepatic ligament, which was divided using Harmonic shears (Harmonic scalpel; Ethicon Endosurgery, Cincinnati, Ohio, USA). Its relationship with the anterior surface of the pancreas was assessed and found no attachment. With gentle traction and countertraction, we could trace the origin of the lesion to be from the lesser curvature of the stomach (Fig. 3). After complete laparoscopic dissection of the lesion until reaching its origin from the stomach, a small portion of the lesser curvature of the stomach to which the lesion was attached was excised using Echelon 60 Endopath stapler (Ethicon Endosurgery). The specimen was placed in an extraction bag and extracted after extending the midline 10-mm port slightly. During surgery, we made every effort to avoid spillage of contents during dissection and manipulation to avoid recurrence. After resection we examined the lesion (Fig. 4). It was a cystic mass measuring about 15 × 10 × 5 cm, multiloculated, thin walled, containing mucin, with some areas of calcification. The patient had an uneventful recovery and she initiated oral intake the night of surgery and was discharged next morning. She was closely followed up by ultrasonography (US)

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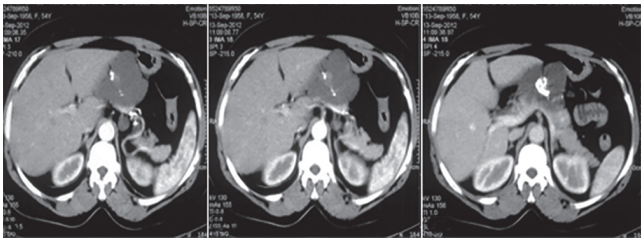
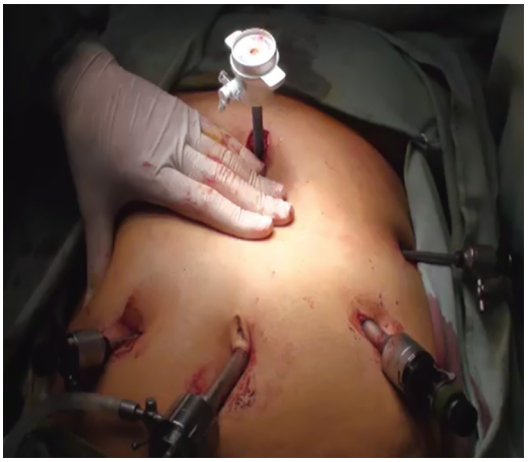
Figure 1



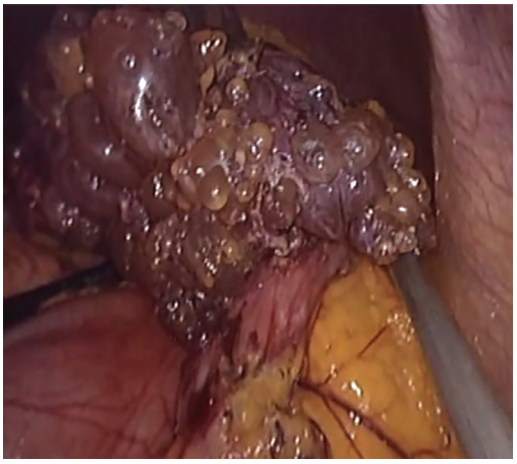
Computed tomographic scan of the patient suggesting pancreatic pseudocyst.

Figure 2



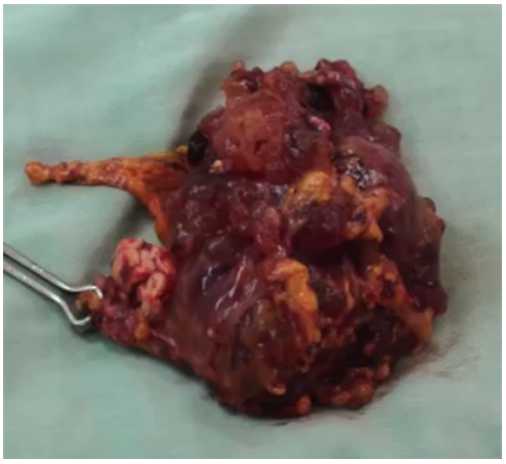
Port design.

Figure 3



After dissection, the tumor was attached by a pedicle to the lesser curve of the stomach.

Figure 4



Tumor after laparoscopic excision.

and CT and remained free of symptoms and had no recurrence for 10 months after surgery.

Microscopic examination revealed multicystic structures lined by flat to cuboidal cells; the wall is fibrous with fibrin deposition and mild inflammatory infiltrate.

Discussion

BMPM, also known as multilocular peritoneal inclusion cysts, is an uncommon lesion arising from the peritoneal mesothelium that covers the serous cavity [3]. It was first identified by Mennemeyer and Smith in 1979 [4]; since then ~150 cases were reported worldwide [5]. Unlike malignant mesothelioma, it is not associated with asbestos exposure [6]. BMPM is characterized by solitary or multiple, thin-walled, multiloculated cysts filled with serous fluid or blood. Their sizes vary from few millimeters to 30 cm [7]. This lesion occurs most frequently in women during their reproductive years [8–10] and is associated with a history of previous abdominal surgery [7], endometriosis [11,12], or pelvic inflammatory disease [7].

Mostly, BMPM is asymptomatic and is often discovered incidentally during examination for other complaints. It may present with vague abdominal pain, nausea, vomiting, and constipation. When it reaches large sizes, it may present as a painful abdominal mass [1]. In addition, its presentation in the form of acute abdomen had been reported [3]. None of the common imaging techniques allow for a definitive differential diagnosis between BMPM and other similar multilocular cystic lesions. In addition, fine-needle aspiration cytology is mostly not specific, and samples show only reactive mesothelial cells. The final differential diagnosis can be obtained from surgical biopsy or through examination of complete surgical specimens [5].

Mesothelioma is a rare tumor arising from the epithelial and mesenchymal elements of the mesothelium, the cellular layer covering most internal organs. The commonest site of mesothelioma is the pleura, followed by the peritoneum and the pericardium [13]. BMPM is also a rare entity with most information coming from individual case reports. It is an intermediate-grade tumor, between benign peritoneal adenomatoid tumors and the more common malignant peritoneal mesothelioma [14].

BMPM mainly involves the pelvic peritoneum, above the Douglas pouch, the uterus, round ligament, spleen, liver, small intestine, appendix, rectum, and previous scars with rare involvement of the upper abdomen and retroperitoneum [15]. A few cases have also been described in the tunica vaginalis [16] and the spermatic cords [17]. The etiology remains unclear and there is no association with asbestos exposure, unlike malignant mesothelioma [6]. Some authors suggest that it is neoplastic, whereas others find it a reactive process. The presence of history of prior surgery, endometriosis, and inflammation suggests that it is a special form of peritoneal reaction to chronic irritation, with mesothelial cell entrapment, reactive proliferation, and cystic formation [3]. Our patient had no history of previous operations. The neoplastic origin is based on slow but progressive growth rates, tendency to recurrence, low incidence of previous abdominal infection, and a high disease-related mortality [8].

The great majority of patients being women of reproductive age suggest a role of female sex hormones in its pathogenesis [18]. BMPM appears typically as solitary or multiple, thin-walled, unilocular cysts filled with serous or mucinous fluid or blood. Their size can vary from a few millimeters to 30 cm. It may be attached or free in the peritoneal cavity. Histologically, the cysts are lined by a single layer of flat or cuboidal cells and mesothelial cells, embedded in fibrovascular stroma. Focal adenomatoid or squamous metaplasia may be seen. Mitosis and atypical cells are usually rare [7]. There have been rare reports of transformation to low-grade malignant mesothelioma, but it is generally classified as a benign process [18]. The gross and histologic findings in our cases were similar to those reported in the literature. Mesothelial cells stain positive for calretinin, thrombomodulin, and cytokeratin 5/6, and these markers allow for a differential diagnosis between BMPM and serum papillar carcinoma of the peritoneum. The cells are negative for endothelial marker CD31 [19].

The differential diagnosis of BMPM includes a variety of malignant and benign lesions that present as solitary or multicystic abdominal masses. Cystic lymphangioma (cystic hygroma), cystic adenomatoid tumor, cystic forms of endosalpingiosis, endometriosis, Mullerian cysts involving the retroperitoneum, and cystic mesonephric duct remnants are the main differential diagnosis among benign lesions. Malignant lesions include malignant mesothelioma and serous tumors of the peritoneum [20]. Multilocular cystic lymphangiomas are the most commonly confused lesions with BMPM. Cystic lymphangiomas usually occur in male children in extrapelvic regions. They are usually found localized to the small bowel, omentum, mesocolon, or retroperitoneum and contain chylous contents. Unlike BMPM, they also have mural lymphoid aggregates and smooth muscle [15].

BMPM usually presents with vague lower abdominal pain, mass, or both, but it is also commonly diagnosed incidentally upon laparotomy for other surgeries [6]. It may also present with obstructive symptoms such as nausea, bloating, or vomiting. In addition, some patients may present with an acute abdomen [3]. Women with this lesion often have a history of prior pelvic surgery, endometriosis, or pelvic inflammatory disease [21]. Physical examination may show abdominal tenderness, distention, or a palpable mass [14]. Imaging modalities that can be utilized include US, CT, or MRI. US demonstrates multiseptated anechoic cysts variable in size and number. The fluid within them is generally anechoic, but they may contain echoes from debris or hemorrhage. CT is often the most useful diagnostic tool as it provides sectional images of the abdominal and retroperitoneal compartments. CT provides information about the location and extent of the mass and demonstrates a well-defined, low-attenuation mass with occasional noncalcified septa. It gives information about relationship with nearby organs, which helps to determine feasibility of resection [22]. MRI provides additional coronal and sagittal planes. The watery serous content has low signal intensity on T1-weighted images and intermediate to high signal intensity on T2-weighted images [23].

Because of its rarity, BMPM treatment options remain an area of controversy. Currently, aggressive surgical resection is the main stay of treatment with palliative debulking and reoperation for recurrence [24]. Aggressive surgical approaches include cytoreductive surgery with peritonectomy and perioperative intraperitoneal chemotherapy to eliminate all gross and microscopic disease [21]. With high recurrence rate and its malignant potential, debulking surgery does not appear to be the most acceptable treatment option for BMPM. Patients may suffer from poorly controlled

chronic abdominal and pelvic pain [24]. There is a 40–55% recurrence rate in female patients and a 33% recurrence rate in male patients [25]. Thus, routine follow-up imaging is required after operation, especially if complete enucleation could not be performed. Because of its rarity, there are no established follow-up or postoperative imaging guidelines. It is recommended to perform follow-up CT scan every 3 months for the first year after operation and then yearly for 5 years due to intraoperative spillage [13]. We followed this follow-up protocol in our patient and the patient remained free of recurrence at 1-year follow-up.

There are few reports of laparoscopic resection for BMPM [26–29], but laparoscopic excision of BMPM of the stomach has not been previously reported in the surgical literature. In our patient, the laparoscopy was undertaken with a presumed diagnosis of a pancreatic pseudocyst. Laparoscopic resection was carried out after verifying the benign nature of the lesion by absence of infiltration to adjacent organs. To ensure a complete excision, the portion of the lesser curvature of the stomach from which the lesion was attached was resected. The use of endo-GIA stapler, not sutures, facilitated this job. Cautious and judicious handling of the tumor is important at laparoscopy to avoid spillage of tumor contents, which is associated with tumor seedlings and recurrence. Laparoscopic surgery for BMPM is far adventitious to open surgery as laparoscopic excision of BMPM does not deprive the patient the benefits of minimally invasive surgery, thus allowing the patient to recover rapidly with minimal pain, and the cosmetic result was excellent.

Finally, preoperative and perioperative recognition of BMPM is difficult but essential for proper surgical management. Although ~150 cases had been described in the literature, publication of more case reports on BMPM is required to better understanding of this entity.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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