BENIGN CYSTIC MESOTHELIOMA OF THE SPLEEN: A CASE REPORT

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ABSTRACT
Benign cystic mesothelioma is a well recognized but rare serosal tumor which mainly arises from the peritoneum in women and is considered as a benign lesion. This is the first case report of benign cystic mesothelioma of the spleen, which took asymptomatic clinical course. A 26-year-old female with morbid obesity and not known to have any medical illness admitted for elective lap sleeve gastrectomy. Ultrasonography was normal except for fatty liver and computed tomography revealed normal result. Laparoscopy was done and during the procedure we found multiple cystic lesion over the anterior surface of the spleen, biopsy was taken from that lesion and the pathology report revealed a benign cystic mesothelioma.

CASE REPORT
A 26-year-old female, not known to have any medical illness, admitted through clinic as a case of morbid obesity for elective lap sleeve gastrectomy. Her body weight was 106 kg, height 160 cm and with BMI 42, other way patient was asymptomatic. On her physical examination patient was hemodynamically stable and afebrile. Her cardiac, chest and abdominal examination was completely unremarkable. The patient's lab investigation which was include: complete blood count, liver, renal function test and electrolyte all were in normal range. Patient went for upper endoscopy as pre-operative assessment and the result was normal except for H pylori which was diagnosed by endoscopy rapid urease test. Ultrasound abdomen was done and it shows fatty liver with normal spleen and gallbladder. CT scan done and it shows normal study. Patient went for lap sleeve gastrectomy surgery and during the procedure incidentally we found multiple small cystic lesions over the anterior surface of spleen (figure 1) and our impression at that time was either metastasis or splenosis. Biopsy was taken from the lesion and the final pathology report revealed Benign Cystic Mesothelioma from the spleen tissue (figure 2).

DISCUSSION
Mesotheliomas are mesenchymal neoplasms originating from the serous lining of the pleural, pericardial or peritoneal space. Multicystic peritoneal mesothelioma involves the peritoneum or extra-peritoneal space, omentum, pelvic or abdominal viscera. Benign cystic mesothelioma of the peritoneum (BCM) was described first in 1979 by Mennemanneyer and Smith [1]. It’s a rare pathological entity with about 130 cases reported in the literature [2, 3]. Several authors consider this tumor as benign [1, 4], and its prognosis is excellent [5]. Although
the peritoneum is the most common origin of this disease, lesions with the same feature can also originate on other serosal membrane. Two cases arising from plural [6, 7], as well as single cases arising in the spermatic cord [8], tunica vaginalis [9], and pericardium [10] have been reported to date. And here apparently we are reporting the first case of the spleen being the only involved organ. The etiology of benign cystic mesotheliomaremand unclear, but it is well known that many inciting factors may promote hyperplastic and neoplastic changes in mesothelial cells. The suggested provoking factors are foreign fibres and dusts, inflammatory mediators, and mechanical injuries [9]. Proliferation and inward migration of peripheral mesothelial cells, proliferation and metaplasia of underlying connective tissue cells, and surface attachment and differentiation of free-floating mononuclear cells all have been postulated as the mechanism of mesothelial cell proliferation in pathological conditions [9]. This peritoneal lesion is characterized by the formation of multiple multilocular thin-walled cysts, which may form large intraabdominal masses [1]. The BCM affects women in 80% of cases, with an average age of 34 years [3]. Diagnosis of benign cystic peritoneal mesothelioma is fraught with difficulties. In many patients, the diagnosis is made incidentally during investigation or surgery for other pathologies, like what happen for our patient. And the clinical presentation is unspecific. It is usually abdominal pain, increased abdominal girth and constipation. Physical examination revealed abdominal distension, abdominal tenderness or a palpable mass [10]. Radiological tests including ultrasonography, CT and MRI may demonstrate the lesions, but cannot differentiate them from other cystic lesions. Fine needle aspiration of the lesion is not informative. Exploratory laparoscopy is the most accurate diagnostic method since it allows local biopsy of the suspected tissue. Pathological differential diagnosis includes a number of benign (cysticlymphangioma, endometriosis and adenomatoid tumors) and malignant lesions (malignantmesothelioma and serous tumors involving the peritoneum). There are no evidence-based treatment strategies for BCM, but surgery with complete enucleation of the cyst to prevent recurrence and possible malignant transformation remains the mainstay of treatment. However, some researchers advocate aggressive surgery followed by heated intraperitoneal chemotherapy (HIPEC) [12]. Indeed, for a long time, the treatment consists of full excision of the lesions (debulking surgery) [7]. Currently, some teams recommend aggressive surgery (extended peritonectomy) followed by HIPEC [3,13]. Two series are available on the results of extended peritonectomy followed by HIPEC. In the first one [13] 5 patients were asymptomatic and 4 showed no recurrence with a follow up between 6 and 69 months. In the second series [14], 5 patients were asymptomatic and 2 had got recurrence with a follow up between 3 and 102 months. The observation of malignant transformation mandates systematic clinical follow-up of these patients. Unfortunately further follow up is compounded by the fact that there are no reliable clinical or imaging feature or tumor markers. The prognosis is excellent, in one of the largest series reported in the literature, only 2 cases of death were reported [10].

Fig 1. Live picture from the operating room showing multiple cystic lesions over the spleen

Fig 2. Histology, Microscopy showing cysts lined with flattened mesothelial cell and the walls composed of loose connective tissue with occasional chronic inflammatory cells

CONCLUSION
Benign cystic mesothelioma (BCM) is a rare tumor with a high local recurrence rate. It requires optimal care in a specialized center especially as there is no evidence-based treatment strategies.

REFERENCES

466


