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CASE REPORT

Low Grade Cystic Mesothelioma Of Rectus Sheath

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ABSTRACT

Low grade cystic mesothelioma or Benign multicystic mesothelioma is a rare neoplasm that arises from the peritoneal surface of abdominal or pelvic viscera as well as extraperitoneal space and omentum. Synonyms like multilocular peritoneal inclusion cysts(MPIC), benign multicystic mesothelioma are used interchangeably in the literature. Multicystic mesothelioma of the peritoneum was first described in 1979. To the best of our knowledge no case of cystic mesothelioma of rectus sheath has been described till date. A case of low grade cystic mesothelioma arising from the rectus sheath in a 35 yrs old female is documented here due to its rarity.

Key words: Benign multicystic mesothelioma, rectus sheath.

Key messages: Low grade cystic mesothelioma should be considered while reporting the cystic masses arising from peritoneal surfaces of areas other than lower abdominal viscera

Introduction

Benign multicystic mesothelioma is a rare neoplasm that involves the peritoneum as well as extraperitoneal space, omentum, pelvic or abdominal viscera. It has a predilection for the serosal surfaces of the ovary, uterus, bladder or rectum [1]. Synonyms like multilocular peritoneal inclusion cysts(MPIC), benign multicystic mesothelioma are used interchangeably in the literature [1]. Multicystic mesothelioma of the peritoneum was first described in 1979 [2] . This tumour has a female predominance with mean age of presentation being 35 yrs [3]. Most common presentation is pelvic and lower abdominal pain , tenderness and/or distension with an abdominal or pelvic mass. They usually have a past history of chronic pelvic irritation resulting from previous pelvic operation, endometriosis and/or pelvic inflammatory disease [3]. Less frequently it may be an incidental finding [1]. Multicystic mesothelioma has no association with asbestos exposure [4].

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This benign entity has a high propensity for local recurrence but no progression to malignancy. [3], [5]. Therefore it is graded as intermediate grade tumour between the adenomatoid tumour of the peritoneum and the more common malignant asbestos-related peritoneal mesothelioma [6]. This neoplasm is neither responsive to chemotherapy nor to radiotherapy [1],[7]. Total surgical excision is the treatment of choice [8]. Histopathological diagnosis is thus very important for management of the patient. Cases of low grade cystic mesothelioma arising from the peritoneal surfaces of pelvic as well as intra-abdominal organs have been reported in the literature [9]. To the best of our knowledge, no case of mesothelioma arising from rectus sheath has been reported till date. A case report of low grade cystic mesothelioma arising from rectus sheath is documented here due to its rarity.

Case Report

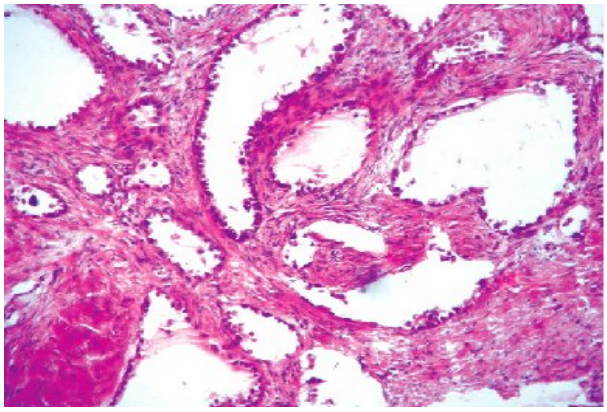
A 35 yrs old female patient presented with the chief complaints of abdominal pain, distension and menorrhagia in the Gynaecology Out Patient Department. She had no significant past medical or

surgical history. On examination an ill defined mass of 6 X 4 cm in size was found in the suprapubic region over the rectus sheath. Routine haematological and biochemical investigations were within normal limit. USG of lower abdomen showed asymmetrical uterine enlargement with heterogenous myometrial echotexture. A markedly heteroechoic mass in the rectus sheath with poorly defined margins and areas of cystic degeneration and septations, suggestive of a resolving haematoma or neoplastic reaction was also noted. Patient underwent hysterectomy during which a mass attached to the rectus sheath was found and a biopsy was taken from the mass. Both the specimens were submitted for histopathological examination.

On gross examination, the uterus was enlarged in size with hypertrophy of the ectocervix. Cut surface showed obliteration of endometrial and endocervical canal. Biopsy tissues (two in number) from the rectus sheath were brown, irregular, firm, measuring 3 X 5 cm each. Cut surface showed multiple minute cystic areas.

Microscopically, uterus with cervix showed adenomyosis uteri with endometrium in proliferative phase and chronic cervicitis. Tissues from the rectus sheath showed multiple cystic areas, lined by mesothelial cells [Table/Fig 1].

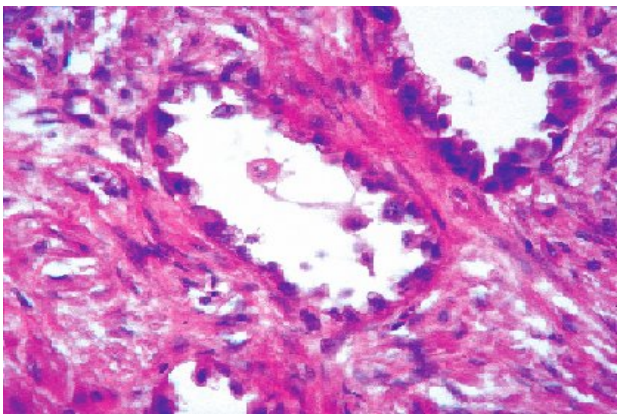
Table/Fig 1



Section from cystic mass showing multiple cysts of varying sizes lined by low cuboidal epithelium (H&E x 40)

In most areas, these cysts were lined by a single layer of cells exhibiting hobnail configuration [Table/Fig 2]. In some areas there were multilayering. Individual cells had vesicular nuclei and prominent nucleoli and some of them showed atypia.

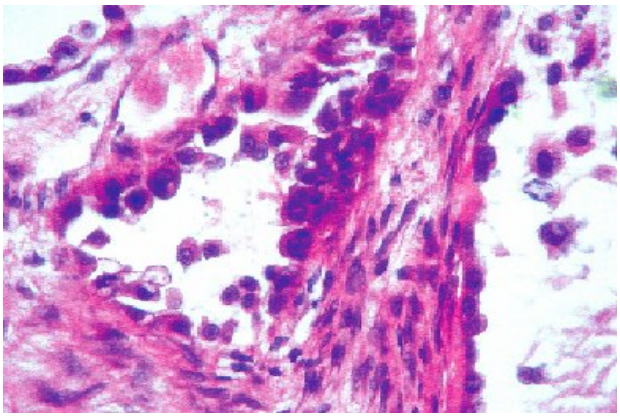
Table/Fig 2



Higher magnification showing 'Hob Nail"appearance of the lining cells.(H&Ex80).

Occasional multinucleated cells were also seen. Lumen of the cyst contained light pink material along with degenerated cells and few inflammatory cells [Table/Fig 3]. Diagnosis of low grade cystic mesothelioma was made. Immunohistochemical staining of the lining mesothelial cells showed reactivity for keratin and calretinin and were negative for CD 31 and Factor VIII related antigen. On the basis of histopathology report the patient was referred to the General Surgery Department for further management. Complete excision of the remaining rectus sheath mass was done. Histopathological diagnosis was again confirmed on the resected mass.

Table/Fig 3



Higher magnification showing mild dysplastic changes of the lining cells.(H&E x120)

Discussion

Benign cystic mesothelioma is a rare lesion arising mainly from the serosal surfaces of ovary, uterus, bladder and rectum [1]. Only few cases arising from other sites have been reported [9],[10]. The case reported here is the only one so far arising from the rectus sheath. Medline search revealed a single case report of mesothelioma in the pleural cavity, three case reports associated with appendix and one case seen in a patient in association with diverticular disease of the large intestine[9],[10]. Two of these three case reports associated with appendix were reported in middle aged women. In the first case, a 53 yrs old female presented with abdominal pain, laparoscopy revealed a [7]. cm retroperitoneal mass close to but not involving the caecal serosal surface [9]. In the second case, a 40 yrs old female patient with the clinical signs of acute appendicitis had a small cystic mass involving the visceral and parietal layers of peritoneum in the appendiceal region [9]. The third one, also presenting with the symptoms of acute appendicitis was a 28 yrs old male, where a 25 cm cystic appendiceal mass was found [9]. The case associated with sigmoid diverticular disease was a 58 yrs old female, ex-smoker, who presented with and was operated for the sign and symptoms of diverticular disease of sigmoid colon [9]. Mesothelioma was an incidental finding. None of the above cases were associated with asbestos exposure [9].

In the present case, a 35 yrs old female patient presented with menorrhagia with an ill-defined suprapubic mass associated with abdominal pain and distension and had evidence of adenomyosis uteri on hysterectomy. However there was no history of previous pelvic surgery or pelvic inflammatory disease. The findings were similar to earlier reports [1],[3] Various differential diagnosis considered were lymphangioma, vascular tumour, other mesenteric –omental cysts, nonpancreatic pseudocysts and endometriosis. Lymphangioma was differentiated by absence of chylous fluid and flattened endothelial lining. Other non specific mesenteric cysts were differentiated on the basis of absence of degenerated material and inflammatory cells. Non-pancreatic pseudocysts usually have thickened wall and endometriosis was ruled out due to absence of endometrial lining and hemorrhage.

Positive immunoreactivity for cytokeratin and calretinin helped in the confirmation of diagnosis as benign cystic mesothelioma whereas negative staining for factor VIII helped in ruling out a vascular lesion. Surgeons reoperated the patient for complete excision of the remaining rectus sheath mass as total surgical excision is the treatment of choice[8]. On further follow up our patient is doing well with resolution of abdominal pain and distension.

The case was considered worth documentation to stress the consideration of this entity while reporting the cystic masses arising from peritoneal surfaces of areas other than lower abdominal viscera.

Conflict of Interest: None declared

References

[1]O'Neil JD, Ros PR, Storm BL, Buck JL, Wilkinson EJ. Cystic mesothelioma of the peritoneum. Radiology 1989; 170:333-337.

[2]Mennemeyer R, Smith M. Multicystic peritoneal mesothelioma: a report with electron microscopy of a case mimicking intra-abdominal cystic hygroma (lymphangioma). Cancer 1979; 44:692-698.

[3]Rosai J. Respiratory tract: nasal cavity, paranasal sinuses, and nasopharynx—larynx and trachea—lung and pleura. In: Rosai J, ed. *Rosai and Ackerman's surgical pathology*. Mosby 2004:2375–6.

[4]V.Kumar, *et al*. Pleural tumours. In: Robbins and Cotran , ed *Pathologic Basis Of Disease*. 7th ed. Philadelphia; Saunders: 2004, p. 768.

[5] Mc Cullagh M,Keen C, Dykes E. Cystic mesothelioma of the peritoneum: a rare cause of 'ascites' in children. Childrens Hospital, Lewisham, London, England.

[6]Weiss SW, Tavassoli FA. Multicystic mesothelioma: an analysis of pathologic findings and biologic behavior in 37 cases. Am J Surg Pathol 1988; 12:737-746.

[7]Bui-Mansfield LT, Kim-Ahn G, O'Bryant LK. Multicystic mesothelioma of the peritoneum. Am J Roentgenol 2002; 178:402 –406

[8]Wendy L. Wong,Theodore A. Johns, ***et al***. Multicystic Mesothelioma. ***Radiographics***. 2004;24:247-250.

[9]Bansal A and Zakhour H D. Benign mesothelioma of the appendix: an incidental finding in a case of sigmoid diverticular disease. J Clin Pathol. 2006;59:108-110.

[10] Ball NJ, Urbanski SJ, Green FH, *et al*. Pleural multicystic mesothelial proliferation. *Am J Surg Pathol* 1990;14:375–80.