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Cystic Adenomatoid Tumour of Uterus – A Rare Entity

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Abstract: Adenomatoid tumour are rare tumours with an overall incidence of 1% in female genital tract. The cystic variant is extremely rare. These tumours occur in both male and female genital tracts and in extragential location like heart, adrenal, appendix, hernial sac, intestinal mesentery, liver, lymphnode, mediastinum, omentum, pancreas, peritoneum, pleura and umbilicus. Adenomatoid tumours are considered to be mesothelial in origin which is proved by immunohistochemistry and ultrastructurally. The histogenesis is being related to entrapment of pleuripotent mesenchymal cells and mesothelial inclusions. This article describes an interesting case of cystic adenomatoid tumour occurring in a 55 years old post menopausal female who presented with abdominal pain. Ultrasonography showed an adnexal lesion arising from left ovary with other possibilities of atypical myoma or intramural haemorrhagic cyst. On macroscopy, the uterus showed a subserosal circumscribed lesion with honey comb appearance. Microscopically the cysts were lined by flattened to cuboidal cells with intervening fibromuscular stroma. Immunohistochemistry showed positivity of these lining cells to HBME-1, Pancytokeratin and focal positivity for Vimentin. Thus to conclude adenomatoid tumour are rare, cystic variant being extremely rare. A strong suspicion of this entity can aid in early diagnosis and excluded other cystic lesions and invasion of a endometrial or a cervical carcinoma when the two co-exists.
Keywords: Adenomatoid, cystic, mesothelial, carcinoma.

INTRODUCTION

Adenomatoid tumour is a rare benign tumour of the female genital tract, most common site being fallopian tube, followed by uterus and ovary. These tumours range in size from 0.2 to 17 cm [1]. As these are smaller in size, they are usually detected incidentally on radiology or after hysterectomy on gross inspection. Most of the cases are difficult to diagnosis on radiology alone, especially the cystic variant. The commoner diagnosis on imaging for cystic lesions include cystic degeneration of uterine leiomyoma, cystic adenomyosis, congenital uterine cysts such as mesonephric and paramesonephric cyst, cervical nabothian cyst, intramyometrial hydrosalpinx and echinococcal cyst [1] hence, a histopathological examination with a high suspicion becomes essential to make the diagnosis of cystic adenomatoid tumour, as these tumours have a benign course without recurrence. Immunohistochemistry using mesothelial markers helps to confirm the diagnosis and exclude the above mentioned differentials which may require further management [2, 3].

CASE REPORT

A 55 year old post menopausal lady presented to the outpatient department with a complaint of pain abdomen and was advised ultrasonography which revealed a left adnexal lesion, arising from ovary with other possibilities of a typical myoma and an intramural haemorrhagic cyst. The patient underwent hysterectomy and on macroscopic examination left adnexae were unremarkable but a subserosal circumscribed lesion with honey comb appearance was noted. The microscopic examination of the subserosal lesion showed multiple, large cystic spaces lined by flattened to cuboidal cells with moderate cytoplasm and round to oval bland nuclei. Intervening stroma was fibrous with admixed smooth muscle fibres at the periphery. Focal patchy lymphocytic infiltrate was also noted. There were no features which would suggest malignancy like pleomorphism, mitoses, necrosis or hyperchromasia. A panel of immune markers were put up to exactly categorize the tumour. The lining epithelial cells showed positivity for Pancytokeratin, HMBE-1 and focal positivity for Vimentin and were negative for CEA and EMA, suggesting a mesothelial origin.

DISCUSSION

Adenomatoid tumours were initially designated as benign mesothelioma of genital tract in 1942 by Masson et al. The term adenomatoid tumor was coined in 1945 by Golden and Ash [3]. The tumour occurs in genital tracts and extragenital sites with an incidence of 1.2% in the hysterectomy specimens [1]. These tumours are usually found incidentally and misdiagnosed radiologically. There are no specific radiological findings to confidently diagnose an adenomatoid tumour, thus highlighting the caveat in the imaging technique.

Fig-1: Gross of the uterus with a subserosal cystic tumor

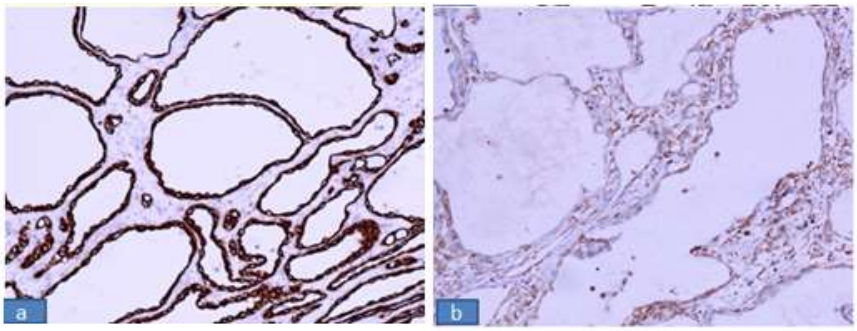

Fig-2: Cystic tumor lined by flattened and cuboidal cells(.a)H &E, X100 and (b)H&E ,X200

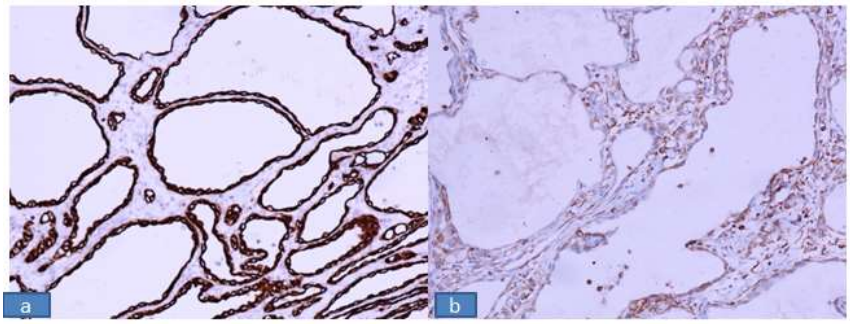

Fig-3: Tumor cells show (a)diffuse and strong positivity for PANCK and (b)focally positivity for Vimentin

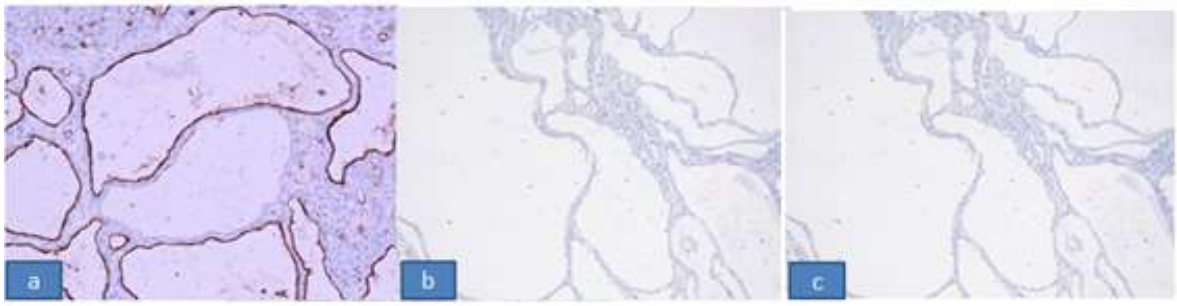

Fig-4: Tumor cells are positive for(a) HBME-1 and are negative for (b)EMA and (c) CEA

The histological examination becomes mandatory to make the diagnosis and exclude other possibilities especially when it has infiltrating pattern and when the tumour is infarcted or when it co-exists with a carcinoma [6, 7]. Macroscopically these tumour can be small solid or large cystic with an average size of 2.1 cm. The later type is the least common and is very rare. A giant cystic adenomatoid tumour has been reported with a size of 15 cm [4]. These cystic tumour shows many differentials radiologically, which include cystic degeneration of uterine leiomyoma, cystic adenomyosis, congenital uterine cysts, such as

mesonephric and paramesonephric cyst, cervical nabothian cyst, intramural hydrosalpinx and echinococcal cyst. Microscopically the tumour can be circumscribed or infiltrating and can be of various patterns, like adenoid, angiomatoid, cystic glandular, solid and tubular. Usually a mixture of patterns is noted but in pure forms they pose a diagnostic difficulty. The cells in few of the cases can be vacuolated, giving signet ring appearance or oncocytic requiring additional diagnostic evaluation. Other histological features identified in these tumour are intratumoural adipose tissue, dystrophic calcification, metaplastic ossification and stromal mucin [3]. Leiomyoadenomatoid tumour a distinct morphological entity which has been reported with prominent smooth muscle proliferation [5]. A study published by Ankur *et al* in 2011, which evaluated the clinicopathological difference of adenomatoid tumour occurring in male and female genital tract and found chronic inflammation to be common in tumours of male genital tract and this has been attributed to the site, more prone to trauma [3].

Adenomatoid tumours can undergo extensive infarction and may co-exist with adenocarcinomas of endometrium and cervix. In infarcted tumour, the diagnostic difficulty is because of necrotic tissue, infiltrating pattern, paucity of typical adenomatoid areas and reactive proliferation of fibroblasts and myofibroblast and in case of co-existent carcinomas the difficulty can be attributed to diffuse infiltrative growth pattern, necrosis, intracytoplasmic, mucin, which may lead to inappropriate surgery. These difficulties can be over come by confirmation of adenomatoid tumour using immunohistochemistry which are positive for mesothelial markers.

The histogenesis of adenomatoid tumours varied from mesothelial in 1940 to a broad spectrum over two decades to endothelial, mesonephric and mullerian [1, 3]. Since then, immunohistochemical and ultrastructural studies have supported mesothelial origin of the tumours. This explains tumours arising in organs which are in proximity to mesothelium. The tumour arising in sites away from mesothelium has seen explained by two hypothesis :-

1. Entrapment of pleuripotent mesenchymal cells.
2. Mesothelial inclusions during embryological development exemplified by adenomatoid tumour in the adrenal gland [8, 10].

These tumour have also been described in HIV patients and renal transplant recipients. Immunosuppression has been implicated to be the cause, like other tumours occurring in the immunosuppressed patients [9, 10].

CONCLUSION

Cystic variant of adenomatoid tumours of uterus are extremely rare and misdiagnosed as leiomyoma, adnexal cystic or other cystic lesions on radiology. A high degree of suspicion is required from the pathologist to accurately diagnose, which can be aided with immunohistochemistry. An associated endometrial and cervical malignancy needs to be exclude in these cases and avoid upstaging the same.

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