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Case Report

Female Adnexal Tumour of Probable Wolffian Origin - A Case Report

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ABSTRACT

Female adnexal tumor of probable Wolffian origin (FAT PWO) is a rare neoplasm wth diagnostic dilemas. It is now considered as a tumor with low malignant potential. Here we share our experience of a case of 65 years old female who was clinically diagnosed as having broad ligament fibroid. But after surgery the microscopic feature simulated Female adnexal tumor of probable Wolffian origin (FAT PWO). We describe here the case along with review of available literature.

Key-words: Female adnexal tumor, Wolffian origin, broad ligament.

Key Messages: Though morphologically benign the female adnexal tumor of probable Wolffian origin should be carefully followed up keeping in mind possibility of recurrence or metastasis.

INTRODUCTION

Kariminejad and Scully in 1973 described nine cases of a distinct extra ovarian neoplasm for which they designated the term "Female adnexal tumor of probable Wolffian (mesonephric duct) origin (FAT PWO) [1]''. These tumors arise within leaves of broad ligment or hang from it or fallopian tube. The tumor are either solid or solid and cystic. Microscopically, they are characterised by epithelial cells growing in cystic structures, solid or hollow tubules and diffuse sheets in sieve –like pattern. Most of the tumors are benign but few cases showed recurrences or metastasis. Here we present a rare case with brief review of literature.

CASE HISTORY

A 65 years old postmenopausal female presented with low abdominal pain. On per vaginum examination a firm mass with irregular surface was felt in right adnexa. Her ultrasonogram showed ill-defined heterogenous mass in pelvic cavity. C.T. scan showed evidence of large calcified adnexal tumour, suggestive of broad ligament fibroid. Her CA-125 levels were within normal limits i.e. 14.8 µ/ml (1.9 – 21 µ /ml).

The patient underwent total abdominal hysterectomy with bilateral salphingo-oophprectomy

In histopathology department we received the specimen of uterus with attached one ovary of 2x1x0.5 cm. Other ovary was of 3x2x0.5 cm with 2 cm long

fallopian tumor with associated mass of size 12x8x8 cm. The mass was capsulated & multi-nodular. On cutting open the mass was solid greyish white with areas of gritty sensation.(Fig 1)

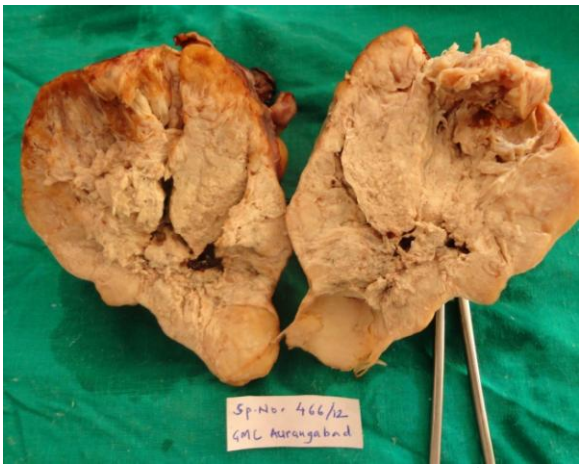

Fig 1: Mass in broad ligament of 12x8x8 cm,Capsulated,bosellated& firm Cut surface - solid greyish white

Microscopically tumor showed well defined capsule with lobules. Lobules showed compactly arranged tubular structure lined by cuboidal lining. Few tubules were dilated. Degenerative changes like hyalinisation, calcification and large areas of fibrosis noted. Both the ovaries and fallopian tubes showed normal histology. (Fig 2,Fig 3)

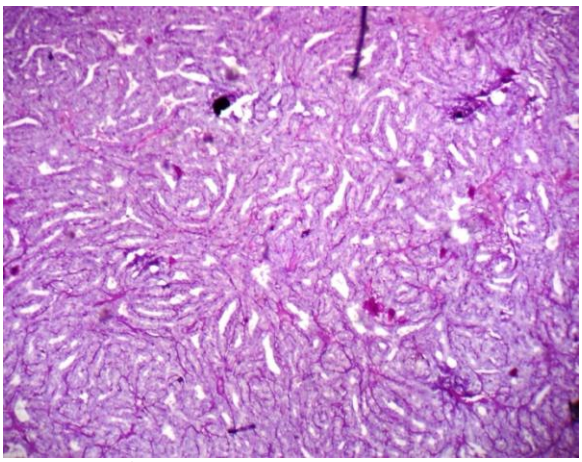

Fig 2: well defined capsule, underneath lobules with compactly arranged tubular structures lined by cuboidal cells with round to oval vesicular nuclei &moderate cytoplasm.(100x)

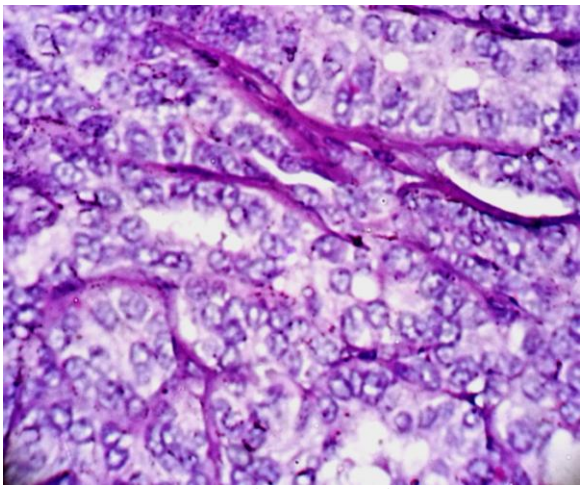

Fig 3: Higher magnification view of tumor (400x)

Immunohistochemically the tumor was positivefor pancytokeratin, vimentin and focally positive for Inhibitin, calretinin. EMA was negative. Considering findings of H & E and immunohistochemistry the diagnosis was kept as female adnexal tumor of probable Wolffian origin.

DISCUSSION

Kariminejad and Scully in 1973 first described nine cases of distinctive female adnexal tumor. [1] They coined the term "Female adenxal tumor of probable Wolffian origin" considering the origin of tumour most likely from Wolffian duct remnants. These tumours were noted within leaves of broad ligaments or hang from it or fallopian tube. As broad ligament regular-ly harbours Wolffian remnants so the tumor origin was suspected. Up-till now about 60 cases are reported in literature. [2] Most cases are seen in broad ligament but cases occurring in rete ovary, retroperitonium & paravagina are also described. [3] Most of the patients present with abdominal pain or mass as in our case. The age of diagnosis ranges from 18- 81 years (average 50 years). [2] In the present case the patient was 65 years old. All of the tumors found to be unilateral and ranged in size from 1 - 25 cm (average 12 cm) in diameter.

The gross appearance of the tumor is solid or sold and cystic, with yellow tan to gray colour. Our patient had multi-nodular gray white solid mass.

The microscopic features were described by Kariminejad and Scully in 1973 as tumors composed of uniform epithelial cells lining cysts and tubules. The cells showed uniform round to oval nuclei and mitoses are rare. The characteristic histologic features of these tumors are closely packed tubular pattern, solid pattern and sieve-like pattern. [3,4]

The diagnosis of FATPWO is further supported by homogenous eosinophilic secretion within lumen of some of the cysts and PAS positive basement membrane surrounding the cysts. The microscopic feature our case resemble to the above mentioned features.

Immunohistochemistry findings are positive for cytokeratin & vimentin were also noted by Irena Sheyn et al. [3] Similarly epithelial membrane antigen was negative in present case.

The differential diagnosis of FATPWO is from sex cord stromal tumor especially Sertoli cell tumor, granulosa cell tumor and Mullerian epithelial tumors. [4,5]

The absence of Leydig cell differentiation excludes sex cord stromal tumors. Carcinomas are excluded by absence of significant cytologic atypia.

These tumors were considered to be benign at the beginning but malignant potential of the tumor has been described.

IrenaSheyn et al reported a 60 years old female with FATPWO who developed liver metastasis after 5 years. [3]

P.E. Hughesdon described a case of ovarian tumor of Wolffian origin of 79years old female with different microscopic patterns that recurred 1 year after initial therapy. [6]

Jerome B.T. and Hector B reported a case of FATPWO in which hepatic metastasis occurred 6 years after resection of the primary tumor. [7] The metastasis displayed more cytologic atypia than the original tumor. Ultra-structurally the tumor consisted of elongated polygonal cells with large irregular nuclei with prominent nucleoli, well developed Golgi apparatus, round to tubular mitochondria, well developed desmosomes and intercellular spaces with microvilli. The ultra-structural features of the tumor did not correspond to those found in normal adult or fetal mesonephric tissue. Still the author concluded that the Wolffian origin of the tumor would not be excluded.

Brescia et al reported an additional case of FATPWO which recurred three times over 16 years period. The patient had one local recurrence and two major metastatic deposits. These were as well differentiated as the original tumor, though the mitotic rate was low and cellular pleomorphism was minimal. The author described the natural history of the tumor as having a tendency for local recurrence as well as distinct metastasis. [8]

Thus, though morphologically benign, the female adnexal tumor of probable Wolffian origin should be carefully followed up keeping in mind possibility of recurrence or metastasis.

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