

# Bilateral Functioning Cystadenofibromas of Ovary

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## Abstract

Cystadenofibroma, a rare variant of benign cystoma of the ovary is generally regarded as a non-functioning benign tumour. Small tumours of this kind are relatively common, but large, bilateral and functioning tumours are extremely rare.

A 48 year old postmenopausal woman presented with bleeding per vagina and abdominal pain. We report a case of bilateral cystadenofibromas of the ovaries, producing oestrogenic substance, in postmenopausal woman.

## Introduction

Epithelial ovarian tumours associated with overt clinical evidence of function are rare.<sup>1</sup> Cystadenofibroma which is a rare variant of benign cystomas of the ovary is generally regarded as a non-functioning tumour. Small tumours of this kind are relatively common, but large fibrous functioning tumours with multiple cysts are extremely rare.<sup>1-10</sup> A case of bilateral cystadenofibromas of the ovaries producing oestrogenic substances is reported. To the best of our knowledge this is the first case to be reported in Indian literature.

## Case Report

A 48 year old postmenopausal woman presented with bleeding per vagina and abdominal pain since 6 months. Per vaginal examination revealed bilateral adnexal masses. USG of the pelvis and abdomen showed bilateral cystic ovarian tumours without ascites and peritoneal implants.

## Pathological findings

*Gross Examination:* Received specimen of total hysterectomy with bilateral salpingoopherectomy; uterus and cervix measured 8 x 5 x 3 cms with unremarkable external surface. Cut section revealed an endometrial broad based sessile polypoidal lesion

measuring 1 x 1 x 0.5 cm and a fundal fibroid of 1 cm in diameter. The cervix was unremarkable.

The right ovarian multiloculated cyst measured 12 x 8 x 4 cms with a grayish white external and internal aspect without any papillary projections or solid areas. The cyst content was serous fluid with the wall thickness of 0.5 cm. The surrounding ovarian parenchyma measured 2 x 2 x 1 cm and showed small cysts. The left ovarian cyst measured 7 x 4 x 4 cms with smooth external surface. Cut section showed a multiloculated cyst with focal papillary projections and a gray white, firm, solid nodule of 2 x 2 x 1 cm. The right and left fallopian tubes were unremarkable.

*Microscopic examination:* Both multiloculated ovarian tumours showed histomorphology of benign cystadenofibroma, consisting of cysts lined by a single layer of cuboidal epithelium with focal stratification and dense fibrous stroma just beneath the epithelium, whereas the stroma elsewhere was myxoid with areas of collagenisation. Focally there were papillary projections which were broad based and lined by cuboidal epithelium. The solid tumour on the wall of the left ovarian tumour had the histology of benign Brenner tumour, composed of clusters of transitional epithelium embedded in a stroma of short fascicles. The endometrial polypoidal lesion showed endometrial hyperplasia with sheets of vacuolated cells in the stroma. The myometrial tumour was a leiomyoma, having interlacing fascicles of spindle cells with cigar shaped nuclei and moderate amount of eosinophilic cytoplasm. The cervix revealed chronic cervicitis with a nabothian cyst.

## Discussion

There is good histological and

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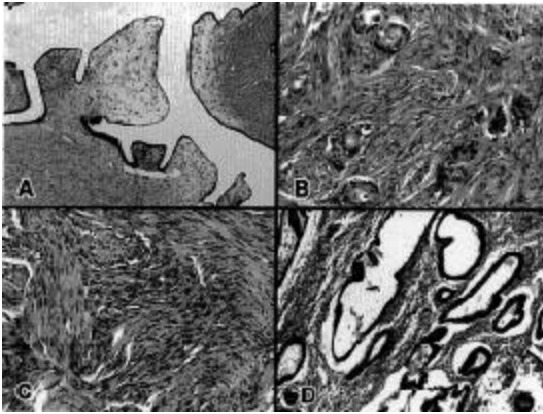


Fig. 1 : Microscopy showing broad based papillae of Cystadenofibroma (A), Brenner component (B), Leiomyoma (C), and Endometrial hyperplasia (D); (H and E x40).

histochemical evidence that stromal cells in epithelial tumours of the ovary are the sites of steroid hormone production. Morris and Scully<sup>5</sup> have described such tumours as “epithelial tumours with functioning stroma”. Ovarian tumours associated with overt clinical evidence of functioning stroma are rare.<sup>1-10</sup> The epithelial tumours which commonly have functioning stroma are mucinous, endometrioid, clear cell and metastatic tumours.<sup>3-10</sup>

The ovarian tumours in this patient were bilateral large cystadenofibromas with Brenner’s component. Cystadenofibromas usually occur in women above 40 years of age with abdominal pain as the commonest presenting symptom followed by vaginal bleeding.<sup>7</sup> We had similar findings in our case.

Grossly, these are usually small, cystic unilateral tumours with clear watery cystic fluid and firm, short rounded papillary projections, which are diagnostic of these tumours.<sup>7</sup> This case had bilateral cystadenofibromas varying from 7 to 13 cm, with characteristic papillary projections present in the left ovarian tumours along

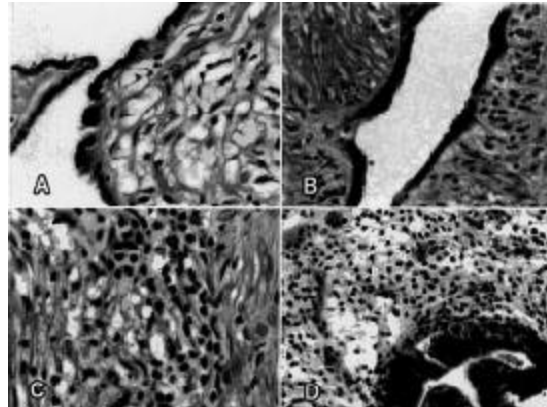


Fig. 2 : Microscopy showing presence of lutein like cells in the stroma of Cystadenofibroma (A and B), Brenner tumour (C), Endometrial hyperplastic polyp (D); (H and E x200).

with a solid area in the cyst wall.

Microscopically these tumours have papillary projections, which are short, broad based structures made of fibrous tissue, and lined by cuboidal ciliated cells. These characteristic microscopic features were present in both the ovarian tumours. Along with this there was condensation of stroma and presence of clusters of polyhedral cells (lutein-like cells) with vacuolated cytoplasm. The histologic evidence of oestrogenic activity in the stroma is condensation and /or luteinization of the stroma just beneath the epithelium.<sup>6,9</sup>

Another approach to establish oestrogenic activity is to examine the end organs affected by these hormones.<sup>4</sup> The end organs affected in this case were endometrium and myometrium with endometrial hyperplasia and leiomyoma respectively. In this regard histological examination and cytologic examination of vaginal smears are the most widely used methods and the findings of active proliferation of hyperplasia in the post menopausal endometrium have been considered suggestive of abnormal high levels of hormonal activity.<sup>4</sup>

The hyperplasia of the stromal cells in the post menopausal ovary containing a non endocrine tumour is a curious phenomenon. Several explanations have been offered.<sup>3-6</sup>

- 1) The mechanical effect of the tumour that expands and compresses the adjacent stroma.
- 2) Ectopic production of hCG or hCG-like substances by the tumour cells.

There is growing evidence that endogenous human chorionic gonadotropin (hCG) is the humoral substance involved in the stimulation of tumour stroma to synthesize steroids.<sup>3-9</sup> This hormone is one of a variety of oncodevelopmental gene products and its production by wide variety of solid non endocrine tumour has been well documented.<sup>3</sup> Immunoperoxidase stains can localize the hCG to the vicinity of the neoplastic epithelium in ovarian tumours, where a high local concentration of this trophoblastic hormone may exist. The frequent localization of the stromal change of luteinization and / or condensation immediately beneath the tumour epithelium could well be or reflection of this local concentration of hCG in that epithelium.<sup>4-9</sup>

Finally the importance of identifying epithelial tumours of the ovary as functioning tumours is stressed, especially in post-menopausal women who present with vaginal

bleeding.

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