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Case Report

An incidental ovarian endometrioid adenofibroma with sertoliform tubules in a patient with endometrial malignant mixed mullerian tumor

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ABSTRACT

Ovarian endometrioid malignancies with sex cord like elements are extremely rare. These neoplasms often get misdiagnosed as sex cord stromal tumors. Although Sertoliform variant of endometrioid adenocarcinoma has been well documented, an endometrioid adenofibroma with sertoliform tubules has not been reported so far in the literature. We report a case of postmenopausal woman with endometrial growth which has been reported as Malignant Mixed Mullerian tumor. She also had an incidental ovarian mass which showed features of an endometrioid adenofibroma with areas resembling sertoli like tubules. This case report is to emphasize the potential of misdiagnosing these entities as sex cord stromal tumors.

1. Introduction

Ovarian endometrioid neoplasms especially carcinomas may rarely resemble sex cord stromal tumors either of granulosa cell or sertoli cell type.[1–3] This has been first described independently by Young and Roth et al in 1982. This morphologic similarity may sometimes be a potential diagnostic pitfall often leading to misdiagnosing these neoplasms as sex cord stromal tumors.

2. Case Report

Patient is a 62-year-old woman who presented with complaints of an episode of postmenopausal bleeding. Per vaginal examination was unremarkable. Imaging revealed a polypoidal lesion in the endometrium. Endometrial curettings was done and reported as high-grade endometri al adenocarcinoma. PET/CT showed disease confined to endometrium with no evidence of nodal spread or distant metastasis. The patient underwent total abdominal hysterectomy with bilateral pelvic and para-aortic lymphadenectomy. She had an uneventful post-operative course. She received chemotherapy thereafter and is currently on followup. The patient is disease free without any evidence of metastases on 1 year followup.

Specimen was sent for histopathologic examination. Gross examination revealed a polypoidal lesion in the endometrium which was solid and cystic. There was no gross myometrial invasion. Left ovary was slightly enlarged with cut surface showing multiple tiny cystic spaces. Other ovary and both fallopian tubes were grossly unremarkable.

On histopathologic examination, the endometrium showed a malignant tumour with varied histomorphologic patterns (Figure 1). One component had papillary, glandular and cribriform architecture with cells exhibiting marked nuclear atypia and numerous mitotic figures morphologically consistent with high grade serous adenocarcinoma. The intervening areas show ed close admixture of dual population of cells with predominant cells being rhabdoid admixed with few scattered adipocytes and lipoblasts. No definite myometrial invasion was noted. Hence a diagnosis of Malignant Mixed Mullerian Tumour with heterologous differentiation was rendered. Sections from

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left ovary showed a neoplasm composed of tubular glands of varying sizes some of which were cystically dilated set in a dense fibrocellular stroma. The tubules were lined by stratified columnar cells some of which were ciliated with endometrioid appearance. Amidst these were some nests, tubules and cribriform pattern with monomorphic population of cells resembling Sertoli cells (Figure 2). The sertoliform cells focally merged imperceptibly with the endometrioid tubules. The cells did not reveal any significant cellular atypia or increased mitosis. The ovary was then all embedded and failed to reveal any evidence of malignancy.

Immunohistochemistry was performed on both endometrial and ovarian lesions (Figures 3 and 4). The glandular complement in the endometrial lesion was diffusely positive for CK 7 and p53. In the stromal component, the rhabdoid cells were diffusely positive for vimentin with patchy positivity for desmin and Myo D1. Adipocytic component was positive for S-100. In the ovarian lesion, endometrioid tubules were diffusely positive for CK 7 and EMA. The Sertoliform tubules were negative for inhibin and calretinin with focal positivity for CK 7 and EMA. Hence a diagnosis of endometrial Malignant Mixed Mullerian tumor with associated ovarian endometrioid adenofibroma with sertoliform tubules was rendered.

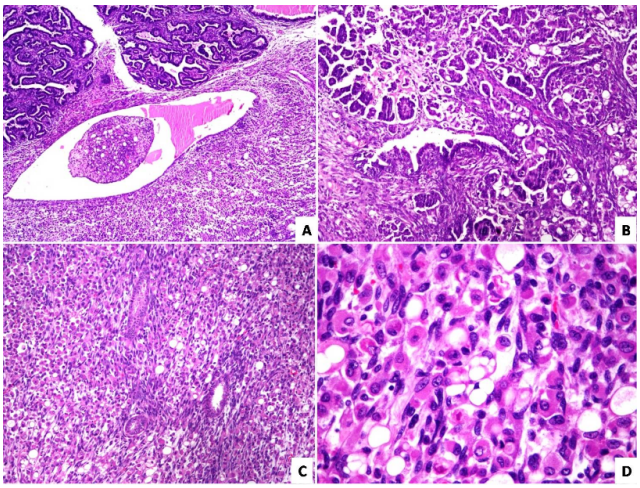

Fig. 1: (A);(H&E-40X) Tumor with two distinct patterns; (B)(H&E-100x)The epithelial component shows features of serous adenocarcinoma (C&D);(H&E-100X &400X): Stromal component shows rhabdoid and adipocytic cells

3. Discussion

Ovarian endometrioid neoplasm with sertoliform tubules are rare and are often misdiagnosed as sex cord stromal tumours. Roth et al was the first to describe the endometrioid adenocarcinoma with sertoliform areas arising from ovary. In their case series, they found that the glandular and sex cord like areas had similar ultrastructural

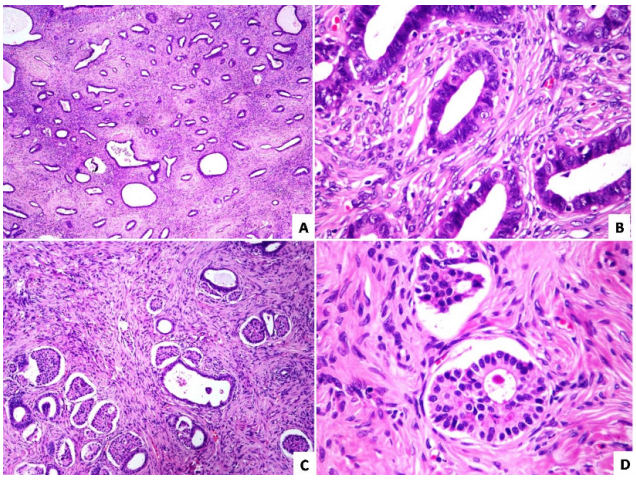

Fig. 2: Endometrioid adenofibroma with tubules lined by endometrioid cells in a fibromatous stroma (A);(H&E-40x) & (B);(H&E-400X). Sertoliform tubules closely admixed with endometrioid tubules (C);(H&E-100x) & (D);(H&E-400X).

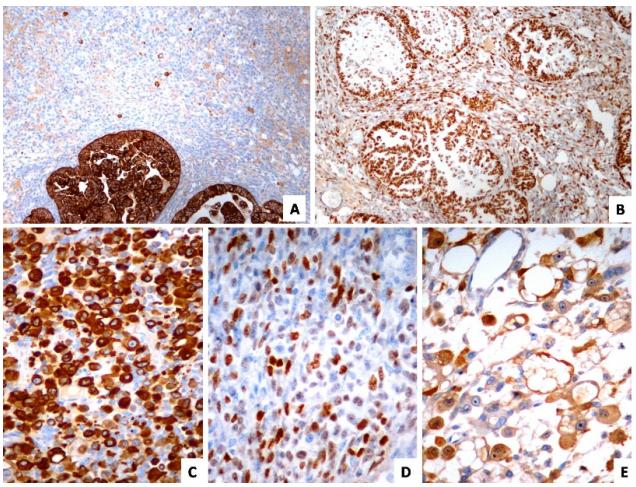

Fig. 3: Epithelial component is positive for CK(A-100x) and p53(B-100x). Stromal rhabdoid cells are diffusely positive for Desmin (C-400x), Myo D1(D-400x) and adipocytic cells are positive for S-100(E-400x)

features, thus suggesting that the two components were indeed different patterns of the same neoplasm.[1]

To the best of our knowledge there are no case reports of a benign endometrioid adenofibroma with sertoliform tubules that are published so far. In our case, the finding that the sertoli like tubules merging with endometrioid tubules along with focal positivity for CK7 and EMA, further supports that these sertoliform tubules may represent a different histomorphologic pattern of the endometrioid neoplasm.

Rarely sex cord stromal tumours may mimic an endometrioid neoplasm. McCluggage et al reported a case series wherein sex cord stromal tumours had tubules

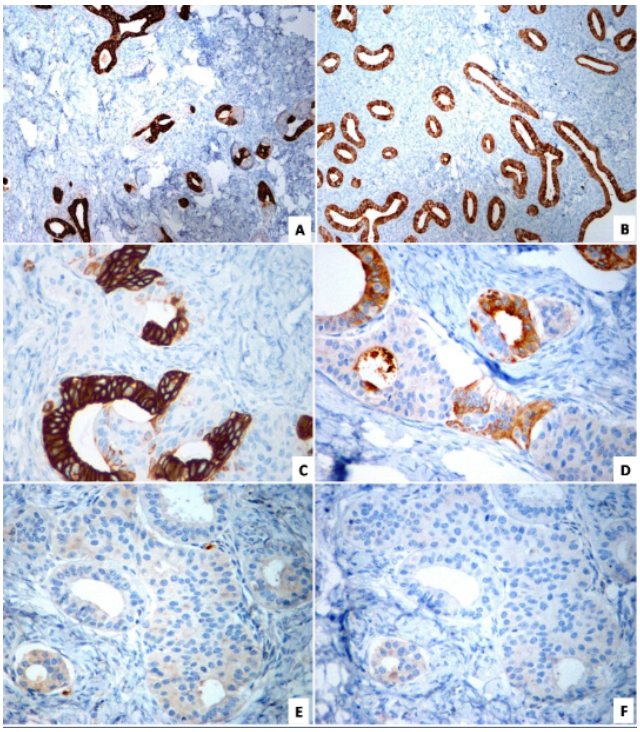

Fig. 4: Endometrioid areas are diffusely positive for CK7(A-100x) & EMA(B-100X). Sertoliform cells shows focal and weak staining with CK7(C-400x) & EMA(D-400x). Calretinin (E-400x) & Inhibin (F-400x) are completely negative.

resembling endometrial glands and were referred to as pseudoendometrioid tubules.[4] These tumours resembled borderline endometrioid adenofibroma or a well differentiated adenocarcinoma. Immunohistochemistry has become established as being helpful in distinguishing these two entities. The pseudoendometrioid tubules in the Sertoli Leydig cell tumour were positive, although focal for alpha inhibin and calretinin and were negative for CK 7 and EMA. The presence of endometriosis or a concomitant squamous metaplasia although not encountered in our case, might provide evidence to the endometrioid nature of the neoplasm.

Endometrioid adenofibroma of ovary coexisting with endometrial endometrioid adenocarcinoma has been reported in the literature[5] and hyperestrogenism has been considered as the causal factor. However the association of endometrial malignant mixed Mullerian tumour with ovarian endometrioid adenofibroma has not been reported so far to the best of our knowledge.

4. Conclusion

This case report is to bring attention to an unusual pattern of endometrioid adenofibroma with Sertoliform tubules. This was a diagnostic challenge as clues to an endometrioid neoplasm such as associated endometriosis and squamous metaplasia were absent in this case. Immunohistochemistry with markers such as CK, EMA, inhibin and calretinin will help us arrive at the correct diagnosis in such difficult cases.

5. Source of funding

None.

6. Conflict of interest

None.

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