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Case Report

Adenosarcoma of uterus in mother and mucinous carcinoma of breast in daughter: a rare case study

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ABSTRACT

Adenosarcoma is an infrequent malignancy which consists of benign glandular epithelium and malignant mesenchymal component. We report a 63-year-old woman diagnosed with adenosarcoma with sarcomatous overgrowth of the uterine corpus, with history of mucinous carcinoma of the breast in her daughter. Although endometrial and breast cancers share few similar hormonal, reproductive and genetic risk factors, the association of endometrial cancer with breast carcinoma is not well established. 63 years old, P4L4, postmenopausal lady presented to our hospital with post-menopausal spotting, foul smelling vaginal discharge and pain abdomen for 1 week. After evaluation she underwent total abdominal hysterectomy with bilateral salpingo-oopherectomy. Intraoperatively, a pedunculated fundal polyp measuring 6×7 cm distending the uterine cavity was noted. Post-operative histopathology examination was reported as adenosarcoma with sarcomatoid overgrowth of the uterine cavity. Immunohistochemistry revealed CK7 (epithelium (+), Vimentin (+), cluster of differentiation 10 (CD10) (+) and, Wilms tumor 1 (-). The possible association between these two conditions, adenosarcoma of uterus in mother and mucinous carcinoma of breast in daughter is explored and presented in this case report.

Keywords: Uterine malignancy, Adenosarcoma, Tamoxifen, Carcinoma breast

INTRODUCTION

Adenosarcoma is an infrequent malignancy which consists of benign glandular epithelium and malignant mesenchymal component. If the sarcomatous part occupies more than 25% of the tumor volume, the situation is referred to as sarcomatous overgrowth – accounting for about 8% of cases. This condition was first described by Clement and Scully.[1] Here in we report a 63-year-old woman who was diagnosed of adenosarcoma of uterine corpus with sarcomatous overgrowth, with history of mucinous carcinoma of the breast in her daughter. Although endometrial and breast cancers share few similar hormonal, reproductive and genetic risk factors, the association of endometrial stromal cancer with breast carcinoma is not well established.[2] The possible association between these two conditions is explored and presented.

CASE REPORT

A 63-year-old postmenopausal female, P4L4, presented to our hospital with chief complaints of post- menopausal spotting, foul smelling discharge per vagina and pain abdomen for 1 week. Per speculum examination revealed a friable growth arising from endocervix which easily bled on touch. Gynecological examination indicated that the uterus was enlarged to the size of 12 weeks, anteverted, bilateral fornices were free and non-tender. MRI was inconclusive about the presence of malignancy.

Interestingly, we found that the 45-year-old daughter of this patient was diagnosed with mucinous adenocarcinoma with neuroendocrine differentiation of right breast and had undergone MRM for the same and was currently on radiotherapy.

After adequate evaluation and informed consent, total abdominal hysterectomy with bilateral salpingo-oopherectomy was performed. Intraoperatively, a pedunculated fundal polyp measuring 6×7 cm in size distending the uterine cavity was noted (Figure 1). Postoperative histopathological examination showed polypoid fragments composed of glandular and stromal components. The glands were cystically dilated and lined by proliferative epithelium. The stroma was highly cellular, especially around the glandular structures with areas of haemorrhage and necrosis and beneath the surface epithelium there was an intraluminal polypoid growth. The stroma showed cellular pleomorphism with frequent atypical mitoses (5-6/10 HPF) (Figures 2 and 3). It was reported as Adenosarcoma with sarcomatoid overgrowth of the uterine cavity.

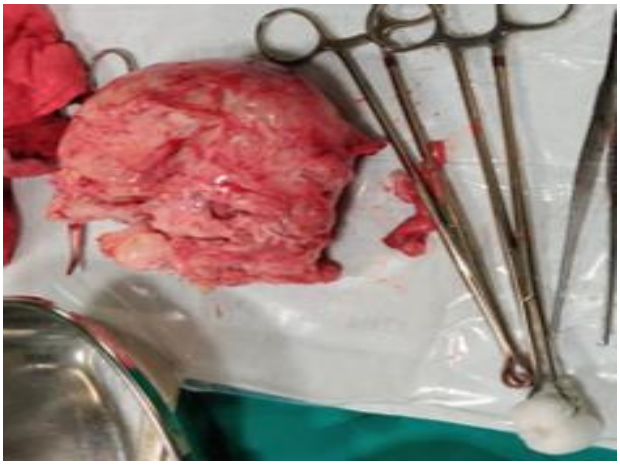

Figure 1: Intra operative findings: bulky uterus.

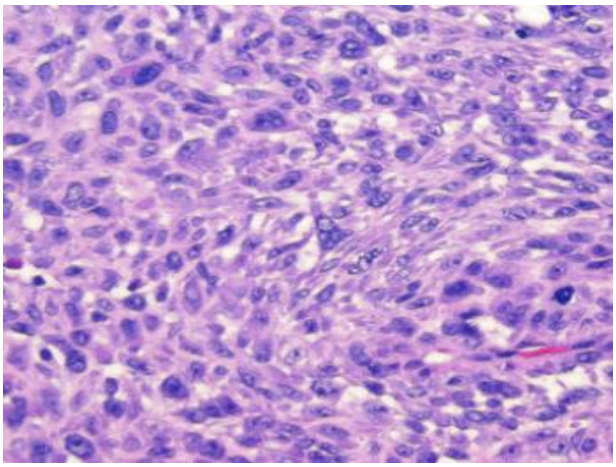

Figure 2: H and E 400X – endometrial stromal sarcoma- The stromal cells show cellular pleomorphism with frequent atypical mitoses.

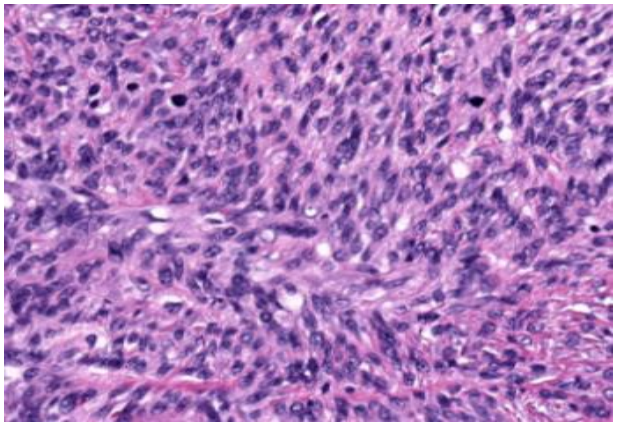

Figure 3: H and E 400X – endometrial stromal sarcoma- The stromal cells show cellular pleomorphism with frequent atypical mitoses.

To confirm the origin of the sarcomatous component, immunohistochemistry was performed. It revealed CK7 (epithelium +), vimentin (+), cluster of differentiation 10 (CD10) (+), Wilms tumour 1 (-) (Figure 4).

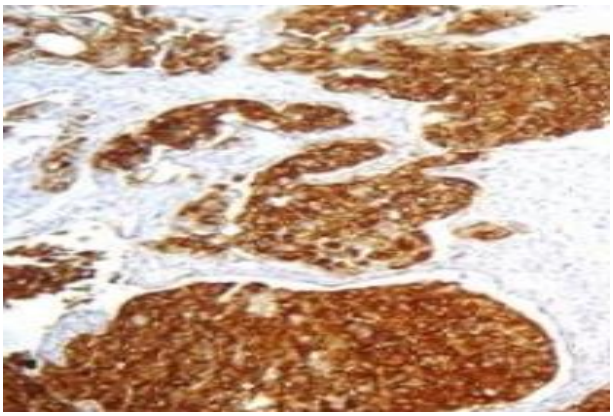

Figure 4: Immunohistochemistry – stromal cells are immunoreactive for CD 10 (strong membranous positivity).

As there was no distant spread, the operative procedure was deemed sufficient and no adjuvant chemoradiotherapy was given. The patient was being followed up regularly, and there is no evidence of recurrence till date (1 year of follow-up).

DISCUSSION

Adenosarcoma of uterus affects women of a broad age range. Incidence is highest in perimenopausal or postmenopausal women but cases have been reported even in childhood.[3] Among the risk factors known to cause adenosarcoma; endometriosis, previous pelvic radiotherapy, long term unopposed estrogen therapy and tamoxifen are well documented. Among these, tamoxifen appears to be more consistently associated with future development of stromal tumors.[4] Sesti et al (2005) have published a case report wherein tamoxifen therapy resulted

in high grade endometrial stromal sarcoma.[5] Liao et al published an article titled "estrogen receptor expression in an endometrial stroma sarcoma after tamoxifen therapy" in 2001. The authors noted that stromal sarcoma can occur after tamoxifen therapy immaterial of status of estrogen receptor expression.[6] Reich et al (2007) published an article titled "Estrogen sulfotransferase (EST) expression in endometrial stromal sarcomas – an IHC study". In their 29-case study, they noted that levels of activity of EST enzyme may have a role in both breast cancer and endometrial stromal cancer.[7] Espinosa et al (2014) have published an article titled "Stromal signatures in endometrioid and endometrial carcinomas" and noted that CSF1 and DTF are stromal signatures identified in breast cancer, hence establishing a link between these two malignancies.[8]

The above-mentioned literature appears to be the only available resources to demonstrate the relationship between endometrial stromal tumors and breast cancer.

Our patient's daughter was a known case of triple negative carcinoma breast and was not on tamoxifen therapy. In case the patient had receptor positive breast carcinoma and needed tamoxifen therapy, caution would have been mandated because she (the daughter) would now be prone for two risk factors – a familial disposition in terms of mother having endometrial stromal sarcoma and taking tamoxifen therapy.

CONCLUSION

The current study mainly emphasizes on the uncommon possibility of association between endometrial stromal tumor and breast cancer leading to therapeutic implications, that is exercise of caution before starting tamoxifen therapy. It also highlights the need for further exploration of biological behavior, tumor microenvironment, for association between endometrial tumors and breast cancers. and clinical prognosis in aiding the patient care and treatment protocols. In brief, where to avoid tamoxifen? is a commonly encountered clinical question. Through this case report, we would like to include women who have a familial predisposition of endometrial sarcomas to the list. Further research is mandated on topic.

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