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Synchronous uterine adenocarcinoma and leiomyosarcoma: A rare case report causing a clinical conundrum

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ABSTRACT

INTRODUCTION: Synchronous gynecologic primary cancers are uncommon. When present, the most frequent malignancies consist of endometrial and ovarian carcinomas. Here we report an exceedingly rare case of concurrent uterine adenocarcinoma and leiomyosarcoma.

CASE PRESENTATION: A 60 year-old female presented with four years of postmenopausal bleeding. An endometrial sampling showed grade 2 endometrioid adenocarcinoma. She proceeded with hysterectomy that contained an anterior endometrial mass and a posterior myometrial mass. The final pathology demonstrated concurrent uterine adenocarcinoma and leiomyosarcoma.

DISCUSSION: To the best of our knowledge, this is the third reported case of simultaneous uterine adenocarcinoma and leiomyosarcoma. As this presentation is infrequent with limited literature, this caused a clinical management conundrum. Unfortunately, the follow-up PET scan suggested possible recurrence or metastasis three months after the surgery.

CONCLUSION: Simultaneous uterine adenocarcinoma and leiomyosarcoma is an exceptionally rare event. As the experience is limited, a multidisciplinary approach in managing these patients may be the best option currently available.

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1. Introduction

Prevalence of multiple gynecologic cancers is only approximately 1% with endometrial and ovarian carcinomas being most frequently observed [1–3]. While concomitant primary malignancies have been encountered for over a century, our understanding of the etiology and management for simultaneous tumors remains unclear. Here we report an exceedingly rare case of hysterectomy and bilateral salpingo-oophorectomy with concurrent uterine adenocarcinoma, leiomyosarcoma and bilateral serous cystadenofibromas.

2. Case presentation

A 60 year-old female smoker with multiple co-morbidities presented with postmenopausal bleeding for four years. Endometrial sampling revealed a grade 2 endometrioid type adenocarcinoma, and the patient underwent total abdominal hysterectomy and bilat-

eral salpingo-oophorectomy. At the time of surgery, there was no evidence of omental or abdominal disease; therefore, no additional staging was performed due to patient's co-morbidities.

Grossly, within the anterior endometrium was a 7.2 cm fungating hemorrhagic mass that invaded the outer half of the myometrium and extended down to the endocervix. Additionally, in the posterior myometrium was a 10.5 cm tan to yellow, soft intramural mass that is completely separate from the anterior endometrial tumor. Within this mass was a softer region (5.1 cm) that contained a focal area of necrosis. The bilateral ovaries were enlarged and cystic (left = 7.1 cm and right = 4.2 cm) and the bilateral fallopian tubes were unremarkable. Incidentally discovered was a 2.2 cm nodule in the right meso-ovarian soft tissue. Histologically, the anterior endometrium was involved by a grade 2 endometrioid adenocarcinoma that invaded 92% of the myometrium and extended to the cervical stromal connective tissue (Fig. 1). The incidentally found meso-ovarian nodule was histologically consistent with metastatic endometrioid adenocarcinoma. Interestingly, the posterior large myometrial mass consisted of predominantly leiomyoma (Fig. 2A). However, within this mass was a region of tumor cells with bizarre pleomorphic nuclei and numerous mitoses including atypical forms (Fig. 2B). Immunohistochemistry was positive for smooth muscle actin and desmin, supporting the diagnosis of leiomyosarcoma. While a diagnosis of carcinosarcoma was considered, the two malignancies involved dif-

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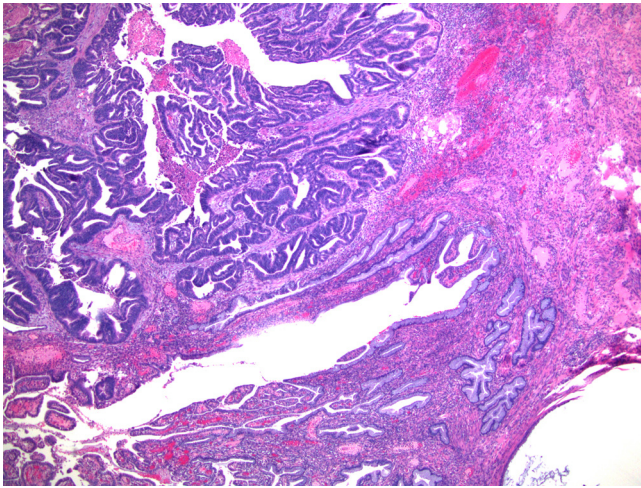


Fig. 1. Endometrial adenocarcinoma (upper left) invading the cervical stromal connective tissue. Benign endocervical tissue can be seen in the lower right.

ferent sides of the uterus and were completely separate from each other; therefore, they were best classified by their own entities. The enlarged bilateral ovaries comprised of serous cystadenofibromas.

3. Discussion

Simultaneous primary neoplasms are uncommon and not well understood. While concurrent endometrial and ovarian cancers are most frequently encountered in gynecologic oncology, only two other cases of concomitant endometrial adenocarcinoma and uterine leiomyosarcoma have been reported in the English literature [4,5]. Sheyn et al. published the first patient with simultaneous adenocarcinoma and leiomyosarcoma of the uterus [5]. A later report described similar findings with the addition of ovarian mucinous adenocarcinoma [4]. Our patient has the two unusual synchronous malignancies previously described with bilateral serous cystadenofibromas.

In cases where epithelial and mesenchymal malignancies co-exist, the more commonly encountered uterine malignancy, carcinosarcoma, has to be considered. Carcinosarcoma accounts for <5% of all uterine malignancies and the diagnosis is made based on a mixture of high-grade carcinomatous and sarcomatous elements in the same lesion. Although our case contained both epithelial and mesenchymal malignancies, grossly these two lesions were located at different sides of the uterus and were histologically distinct from each other. For these reasons, two different primary diagnoses were rendered. This is an important distinction as clinical management for carcinosarcoma would differ.

Several studies have suggested that synchronous endometrial and ovarian primary cancers have an earlier stage and a lower grade disease at the time of presentation, with a 25% reduction in risk of death when compared to patients with single tumors [6–8]. Unfortunately, this improved prognosis cannot be extrapolated for this case as experience in this rare presentation is limited in the literature. Optimal treatment course for synchronous malignancies is controversial given different regimens would be considered depending on cancer types and stage [6]. In this case, the leiomyosarcoma is stage IB, as it is limited to the uterus and the mass itself is over 5 cm. Clinical management would include observation and possible chemotherapy such as gemcitabine and docetaxel. Staging for endometrial adenocarcinoma in this patient is incomplete given full staging was not performed due to her co-morbidities. However, the presence of metastatic adenocarcinoma involving the meso-ovarian soft tissue would be considered at least stage IIIA, which would lead to radiation and chemotherapy including agents such as paclitaxel and carboplatin. This chemotherapeutic regimen differs if considering the diagnosis of carcinosarcoma, in which case treatment includes ifosfamide in combination with paclitaxel or cisplatin. Unfortunately, three months post-surgery, the patient had a PET scan showing abdominal and pelvic retroperitoneal lymphadenopathy, suggesting potential metastasis. While the primary source of the metastasis is uncertain, given her high-stage endometrial cancer, adjuvant chemotherapy with cyclophosphamide, adriamycin and cisplatin was recommended [9].

Simultaneous uterine adenocarcinoma and leiomyosarcoma is an exceedingly rare event. Given the uncommon nature of this presentation, it is difficult to know the exact disease course and management for these patients. In this rare scenario, a multidisciplinary approach is extremely helpful as discussion with clinicians with various expertise can formulate the best treatment options for this patient.

Conflict of interest

The authors have no relevant financial conflicts or conflicts of interest to disclose for this work. Dr. Alvarez reports personal fees from Tracon Pharmaceuticals, Inc. outside the submitted work.

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None.

Ethical approval

UC Davis IRB oversight was obtained.

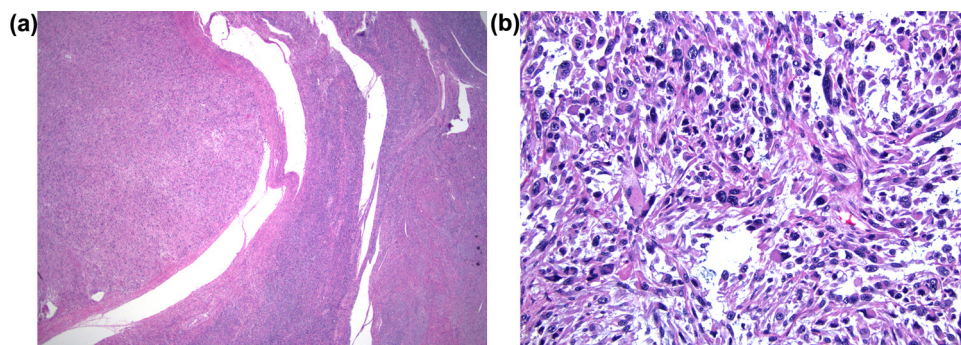


Fig. 2. (A) Low power image of leiomyosarcoma (left) adjacent to leiomyoma (right). (B) High power image showing pleomorphic spindle cells with bizarre nuclei and mitoses with atypical forms.

Consent

Consent was unable to be obtained as patient and all contacts were unable to be reached. IRB approval was obtained and consent waiver provided.

Author contribution

Katie K. Crean, M.D. – literature review and writing the manuscript.

Eric C. Huang, M.D., Ph.D. – case report concept, writing and providing images for the pathology portion of the manuscript, manuscript revision.

Edwin A. Alvarez, M.D. – case report concept, manuscript revision.

Guarantor

Edwin Alvarez, M.D.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <http://dx.doi.org/10.1016/j.ijscr.2016.03.005>.

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