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## Correspondence

## Serous carcinoma arising from adenomyosis



Adenomyosis (variant diseases of the endometriosis) is still a biggest challenge for both physicians and patients [1–3]. Common symptoms and signs include an elevated of serum level of CA 125, dysmenorrhea, menorrhagia, and infertility [4,5]. Because of poor understanding of pathophysiology of adenomyosis [6], the current treatment strategy might be only limited to symptom treatment, including hormone therapy to induce menopause status, waiting for spontaneous menopause, and the destructive surgery, such as total hysterectomy and/or adenomyomectomy, cytorreduction of adenomyosis. The therapeutic responses are varied greatly. Although organ-preservation has been getting more popularity, the risk of concealed malignancy of adenomyosis and possible adenomyosis-associated malignancy, such as endometrial cancer is never overlooked. Furthermore, the increased risk of malignancy still continues, even though these women have been after menopause [7]. The following case reports a postmenopausal woman who had an unexpected diagnosis of serous carcinoma arising from adenomyosis.

A 67-year-old woman complained of the right inguinal mass lesions and postmenopausal spotting. Transvaginal ultrasound showed a cystic mass lesion within the myometrium area on the posterior uterus, and 4 mm in thickness of the endometrium. Papanicolaou smear showed negative findings. Then, she underwent a dilatation and curettage for postmenopausal bleeding and ultrasound-guided biopsy for inguinal masses. Pathology revealed a metastatic serous carcinoma of inguinal lymph node biopsy but a negative finding for a dilatation and curettage. Then, serial examinations were performed, including whole body positron-emission computed tomography (PET-CT), which showed abnormal findings on the uterus and bilateral inguinal areas. Based on the diagnosis of cancer of unknown origin but highly favoring uterine origin, the woman received a laparoscopic complete staging surgery and inguinal lymph node dissection. Final pathology showed serous cancer arising adenomyosis and the entire endometrium was intact without identified cancer. Pelvic lymph node was also negative for malignancy but bilateral inguinal lymph node showed metastases. Then, the woman received postoperative chemotherapy using a combination regimen of paclitaxel and cisplatin and now she is free of disease for 8 months.

This case is interesting and worthy of discussion. First, the diagnosis of cancer arising from adenomyosis is difficult and often delayed because of the absence of tumor in the eutopic endometrium. Our current presented case was also negative for transvaginal ultrasound screening (4 mm in thickness) and then negative for dilatation and curettage finding.

Second, cancer arising from adenomyosis is often endometrioid type (76.1%) [8], which is still significantly lower than that of endometrioid-type endometrial cancer coexisting with adenomyosis (85.2%) or all endometrial cancers (>80%) [8,9]. In addition,

cancer arising from adenomyosis presents a more aggressive clinical behavior and worse outcome (decreased disease-free survival with adjusted-hazard ratio 2.87 and 3.07, 95% confidence interval 1.44–5.70 and 1.55–6.08, respectively) [8,10]. By contrast, the prognosis of patients with endometrial cancer coexisting with adenomyosis is often better than patients with pure endometrial cancer without adenomyosis [10]. The conflicted results might make the audience confused.

To further clarify the outcome of serous cancers arising from adenomyosis, we used the term “adenomyosis and serous cancer” (from 1956 to May 20, 2017) to search PubMed for relevant English-language articles (<https://www.ncbi.nlm.nih.gov/pubmed/?term=adenomyosis+and+serous+cancer>) and identified only a few cases [7–12]. According to literature review, Mutsuo's group might be the biggest series to investigate the cancer arising from adenomyosis [8,10]. Mutsuo and colleagues used a systematic literature search to identify 46 cases with cancer arising from adenomyosis [8,10]. Based on their study and our further extensive literature review [7–12], the reported cases for serous cancer arising from adenomyosis without endometrial involvement are only 7 (2 cases in the Lu's study [7], 2 cases in the Abushahin's study [11], 1 case in the Izadi-Mood' study [12], 1 case in the Koshiyama's study [13], and one current case in ours). The outcomes seemed to be worst in the literature review.

Third, the strategy in the management for women with serous cancer arising adenomyosis is extremely limited. We believed that we can use the similar strategy which has already been applied to women with uterine papillary serous cancer [14] or far-advanced pure endometrioid endometrial cancer [15]. This strategy contains two critical components. One key step is complete resection of the tumor, which includes a complete and thorough lymphadenectomy. The second key step is the use of postoperative paclitaxel-based multiple-agent chemotherapy with and/or without radiotherapy. Combination of an aggressive and extensive surgical approach and following paclitaxel-based chemotherapy might have a chance to provide a better survival for those patients with serous cancer arising from adenomyosis, similar to our above-mentioned case in the current report.

In summary, there are two key messages in our present case. First, it is difficult to make a preoperative diagnosis of serous cancer arising from adenomyosis. An extensive, complete and thorough surgical excision and following postoperative paclitaxel-based chemotherapy should be applied to all patients diagnosed with this highly aggressive disease.

## Conflicts of interest

All authors declare no conflict of interest.

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