



## Serous Adenocarcinoma of Endometrium Coexistence with Atypical Polypoid Adenomyoma in a Postmenopausal Woman: A Rare Case

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This work describes a rare case of serous carcinoma coexistence with atypical polypoid adenomyoma (APA). A 55-year-old postmenopausal woman presented with intermittent vaginal bleeding for 1 year. Her Pap test exhibited atypical glandular cell and endometrial biopsy showed serous carcinoma. She underwent a staging surgery, and the histopathology revealed serous carcinoma with p53 mutation and coexistence with APA staged as FIGO Stage Ia, Grade 1. The patient recovered well postoperatively. APA lesion is generally benign, but it may progress in rare cases to adenocarcinoma, usually endometrioid type. This is the first case report regarding serous carcinoma coexistence with APA. Therefore, the patient with APA should always be closely followed up. Nevertheless, further study is stipulated to better understand the clinicopathological features of serous adenocarcinoma coexisting with APA.

**Key words:** Atypical polypoid adenomyoma, endometrium, menopause, serous carcinoma

### INTRODUCTION

Atypical polypoid adenomyoma (APA) is a rare uterine lesion first described in 1981.<sup>1</sup> APA generally occurs in premenopausal women, and it involves the endometrium of the lower uterine segment. Fertility preservation is a common issue among women who are still fertile. APA is considered as a benign lesion, which may recur or persist following conservative treatment with polypectomy or curettage. Furthermore, synchronous or metachronous occurrence of atypical hyperplasia or endometrioid adenocarcinoma within APA has been reported.<sup>2-4</sup> Nevertheless, serous carcinoma coexisting with APA has never been reported up to now. This study reports a rare case of serous adenocarcinoma coexistence with APA in a postmenopausal woman.

### CASE REPORT

A 55-year-old Chinese woman (gravida 2, para 2), with a body mass index (BMI) of 33.8 kg/m<sup>2</sup>, presented in 2013 with intermittent vaginal bleeding for 1 year. Her menarche occurred

at 14, and her menopause was at the age of 42. Although she was followed up regularly for 1 year, no abnormal finding was observed. Her vaginal ultrasonography showed thickening endometrium in 2014. Pap test at this visit revealed atypical glandular cells. Dilation and curettage was then performed, and the pathology revealed serous carcinoma. Tumor makers of cancer embryonic antigen, squamous cell carcinoma, CA 125, and CA 19-9 were 0.81 ng/ml, 1.9 ng/ml, 10.8 IU/ml, and <1.2 IU/ml (all values within normal limits), respectively. Subsequently, she underwent staging surgery including total abdominal hysterectomy, bilateral salpingo-oophorectomy, and bilateral pelvic lymph node dissection.

Grossly, the uterus was slightly enlarged measuring 10.5 cm × 7.5 cm × 3.7 cm in size. The serosa was glistening and smooth. On cutting, the endometrial cavity was 5.2 cm × 3.6 cm × 2.7 cm in size, and the endometrium was 0.5 cm in thickness. An obvious pedunculated polypoid lesion measuring 6.4 cm × 2.7 cm × 1.7 cm was seen [Figure 1a]. In addition, two protruding mucosal lesions were also

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found [Figure 1b]. The myometrium was measured up to 1.8 cm in thickness. The endocervical canal was 4.5 cm in length. Bilateral ovaries and tubes were grossly unremarkable.

Microscopically, the first lesion exhibited serous carcinoma located adjacent to APA [Figure 2a]. Haphazardly arranged irregular glands were found deep in a smooth muscle stroma [Figure 2b]. The serous carcinoma was continuous with APA [Figure 2c] and positive staining of p53 [Figure 2c]. The other two lesions showed benign endometrium polyps. The tumor was staged as Stage IA, Grade 1 serous adenocarcinoma of endometrium as defined by the International Federation of Gynecology and Obstetrics classification.

She recovered well postoperatively, and she showed no evidence of disease for 1.5 years.

## DISCUSSION

APA, described as a benign polypoid lesion of the uterus, is an uncommon endometrial lesion.<sup>1</sup> Therefore, conservative management with polypectomy or curettage may cause recurrence or persistence in 30–45% of patients. The recurrent risk is lower when treated with transcervical resection (TCR, 10%) than with dilation and curettage (36.4%). Nevertheless, careful follow-up is stipulated because of the risk of recurrence of APA or transition to endometrioid adenocarcinoma.<sup>5</sup> Coexistence of endometrioid adenocarcinoma with APA has been reported as 17.2%.<sup>6</sup> However, no case reports have been observed of serous adenocarcinoma coexistence with APA.

APA of low malignant potential has been proposed to have a higher rate of recurrence and progression to carcinoma than typical APA.<sup>7</sup> The dilation and curettage specimen obtained during the initial treatment in this case showed serous adenocarcinoma. The hysterectomy specimen showed serous adenocarcinoma in continuity with APA, indicating coexistence of APA with serous carcinoma.

APA is a biphasic neoplasm with epithelial and mesenchymal components. The hyperplasia or EC may

involve the APA.<sup>2–4</sup> Serous adenocarcinoma of endometrium is a Type 2 endometrium cancer, and it is widely considered to develop from endometrial intraepithelial carcinoma, which is related to malignant transformation of the endometrial surface epithelium (such as a benign endometrial polyp).

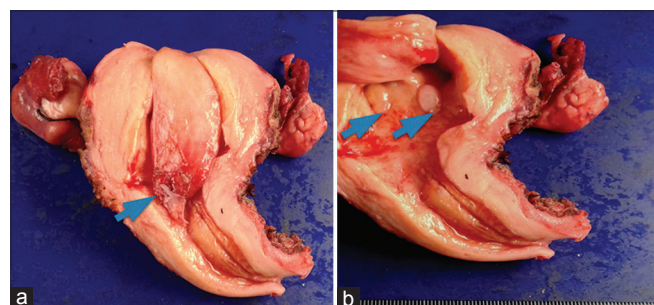
Type 2 endometrial cancer is conventionally considered to differ from Type 1 tumors in several ways. First, the average age at diagnosis is older for the Type 2 disease than for the type 1.<sup>8</sup> Second, serous adenocarcinoma is not related to estrogenic stimulation, and most cases have p53 mutations.<sup>9</sup> Third, obesity is a known risk factor for Type 1 endometrial carcinomas, but was not traditionally associated with Type 2.<sup>10</sup> Fourth, women with Type 2 tumors are more likely to be parous than nulliparous.<sup>11,12</sup> This case is a 55-year-old, obese (BMI 33.8 kg/m<sup>2</sup>), parous, and postmenopausal woman.

TCR has been recommended for the diagnosis and treatment for patients who wish to receive conservative treatment for APA.<sup>6</sup> Careful follow-up with ultrasonography, hysteroscopy, and endometrial biopsy should be performed, although appropriate examination intervals have not been determined clearly.<sup>5</sup>

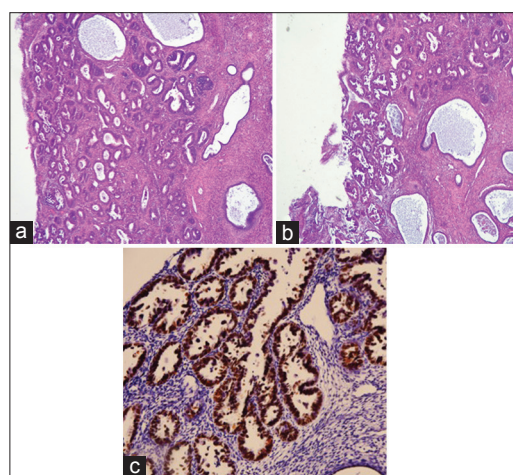
Medical professionals consider serous carcinoma to be highly aggressive and more advanced than endometrioid carcinoma.<sup>13</sup> This patient was staged as IA, Grade 1 at the time of diagnosis. The patient was treated with staging surgery. No recurrence was observed 5 months postoperatively.

## CONCLUSIONS

Physicians should be aware that APA may coexist with serous adenocarcinoma. Therefore, close follow-up should



**Figure 1:** Gross picture of the uterus and tumor. (a) A pedunculated polypoid lesion filled the upper segment of endometrial cavity. An area of reddish discoloration on the tip of this polypoid lesion (arrow). (b) Gross appearance of two endometrial polyps (arrows)



**Figure 2:** Histology of the tumor. (a) Histology of atypical polypoid adenomyoma was characterized by haphazardly structured irregular glands in a smooth muscle stroma (H and E, ×40). (b) Serous carcinoma continuous with atypical polypoid adenomyoma (H and E, ×40). (c) Positive p53 immunostaining of serous carcinoma (×200)

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be performed for a patient with APA if she desires fertility. Furthermore, treatment with staging surgery is mandatory if cancer was detected on APA. Nevertheless, further study is required to better understand the clinicopathological features of serous adenocarcinoma coexistence with APA.

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### Conflicts of interest

There are no conflicts of interest.

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