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Case report

Endometrial adenomyoma polyp caused postmenopausal bleeding mimicking uterine malignancy

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ABSTRACT

This case report presents a 66-year-old postmenopausal woman with a case of endometrial adenomyomatous polyp (EAP) that presented as postmenopausal vaginal bleeding and mimicked endometrial cancer. The ultrasonography revealed a mildly enlarged uterus approximately 7.1 cm × 3.7 cm in size. The endometrium was 1.9 cm in diameter. The findings of magnetic resonance image (MRI) comprised abnormal intrauterine lesions with multiloculated cystic components. Endometrial biopsy by Pipelle was performed, and revealed hematoma. The hysteroscopy was then arranged, and two polypoid tumors were found. Tumor resection was performed, and the histology of the tumor was adenomyoma. EAP is a rare benign tumor of the uterus that is not easy to differentiate from endometrial cancer by ultrasound or MRI. Hysteroscopy is recommended when the results of tissue sampling by Pipelle differ from the image findings.

Introduction

Although endometrial polyps are commonly detected, endometrial adenomyomatous polyps (EAP) are rarely detected. A previous study revealed that EAPs account for 1.3% of endometrial polyps.[1] Endometrial polyps are usually asymptomatic. However, large endometrial polyps may present with abnormal uterine bleeding. The standard treatment for endometrial polyps is hysteroscopic resection. Hysterectomy is also the treatment of choice for large adenomyomatous polyps mimicking leiomyosarcomas.[2]

The World Health Organization 2014 classifications of adenomyoma include atypical polypoid adenomyoma (APA), endometrial-type adenomyoma (EA) and endocervical-type adenomyoma.[3,4] APA is composed of endometrial glands embedded in a myomatous stroma[5] and has a benign behavior. H-caldesmon and CD10 are used to distinguish APA from endometrioid adenocarcinoma.[3] EA is also composed of endometrial-type glands surrounded by mature smooth muscle (the predominant component) and may be polypoid or intramyometrial.[3] EAP may be confused with APA, however, the latter has a prominent glandular component. Endocervical-type adenomyoma is uncommon and is composed of mucinous glands embedded in abundant smooth muscle.[3]

We report the case of a woman who presented with a large submucosa type of EAP, complicated with postmenopausal vaginal bleeding and no diagnosis following endometrial biopsy. Subsequently, hysteroscopy found a large EAP which was resected.

Case Report

A 66-year-old woman had undergone hormone replacement therapy 3 years before presenting with postmenopausal vaginal bleeding. She had a Stage IV cystocele with mesh implantation 3 years previously. She received a Pap smear in the previous 3 years with normal findings. Owing to the development of postmenopausal vaginal bleeding, she visited our outpatient department (OPD), where the gynecological examination revealed vaginal bleeding without lifting pain or palpable adnexal mass. The pelvic ultrasonography revealed an enlarged uterus, approximately 7.1 cm × 3.7 cm in size. The thickened endometrium was approximately 1.9 cm in diameter (Figure 1). The magnetic resonance image (MRI) revealed abnormal intrauterine lesions with multiloculated cystic appearance and soft tissue components (Figure 2).

Conflicts of interest: All contributing authors declare no conflicts of interest.

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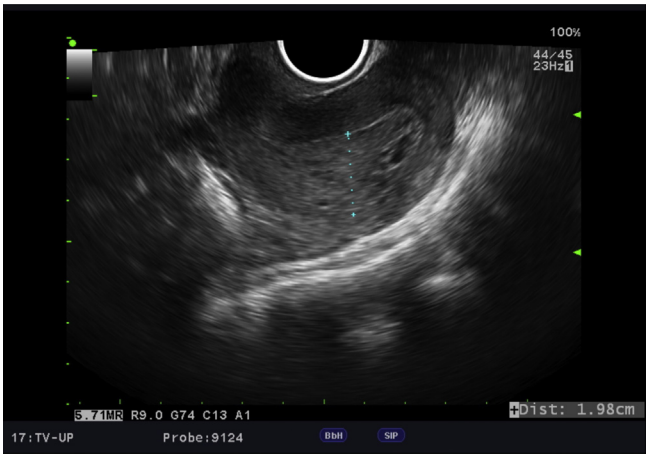

Figure 1. Transvaginal ultrasonography revealed a complex heterogeneous tumor in endometrial cavity. The largest tumor thickness was 1.98 cm.

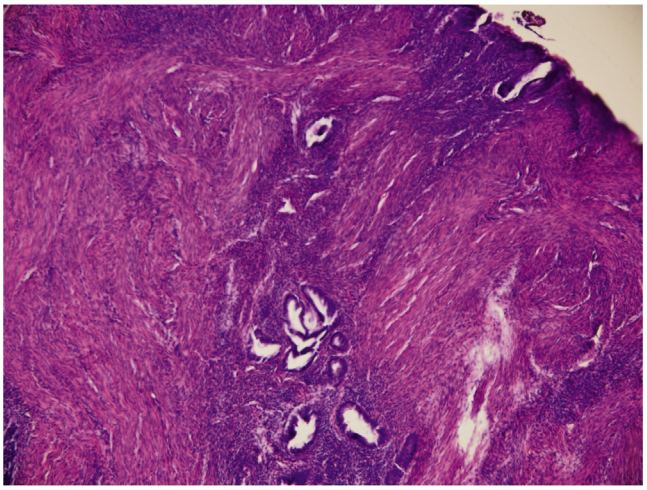

Figure 4. Histopathology of the adenomyoma. Admixture of endometrial-like glands, intersected by fascicles of smooth muscle bundles (Hematoxylin and eosin staining, 100×).

No pelvic lymphadenopathy was noted. Furthermore, the cancer antigen 125 was 10.1 IU/mL (within normal limit), and normal complete blood count and biochemistry tests were noted. The endometrial biopsy by Pipelle (Endocurrette; Midvale, UT, USA) was performed at the OPD, and the pathology revealed hematoma. Hysteroscopy was arranged after discussion with the patient. The hysteroscopy revealed two polypoid tumors with uneven and hemorrhagic surfaces, located at the uterine fundus and posterior wall (Figure 3). Transcervical tumor resection was performed thereafter. Histology of the polypoid tumors revealed adenomyosis (Figure 4). No bleeding was noted 1 week following tumor resection.

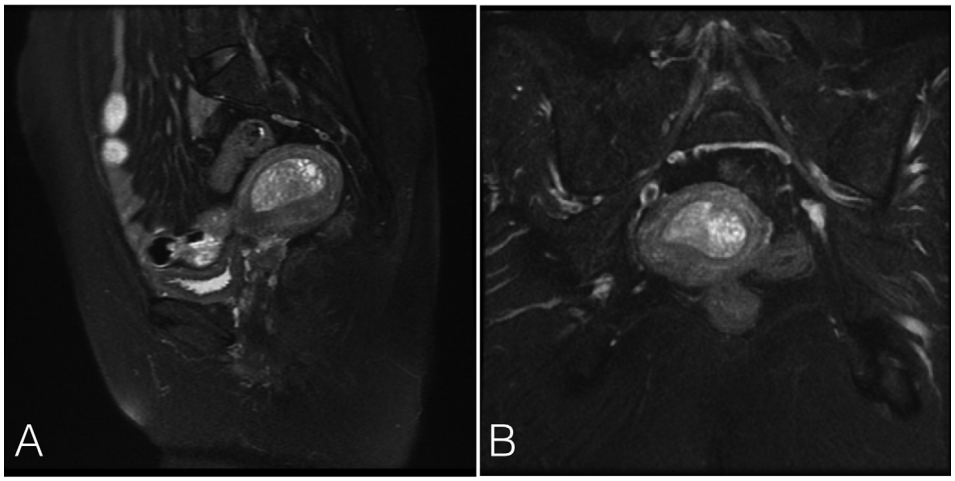

Figure 2. Magnetic resonance image of the adenomyoma showing an endometrial mass. (A) Sagittal T2-weighted image; (B) transverse T2-weighted images. Endometrial tumors have a multiloculated cystic appearance and soft tissue components.

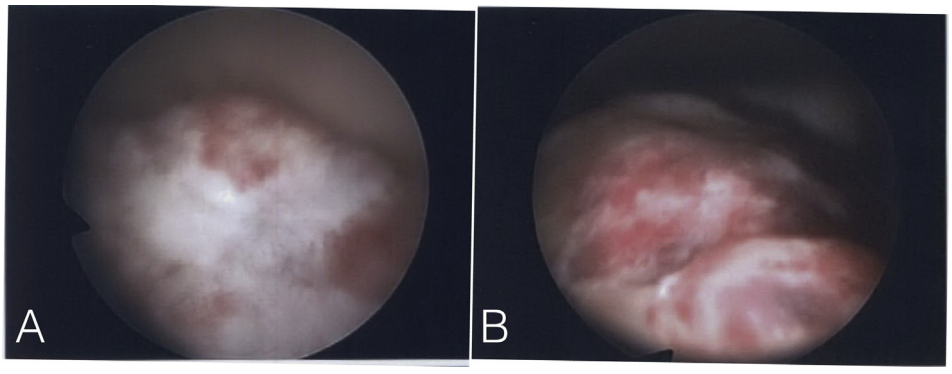

Figure 3. Hysteroscopic view of adenomyoma. (A) and (B) different aspects of adenomyoma. Uneven and hemorrhagic surface of the polypoid tumor was noted.

Discussion

Vaginal bleeding may be the initial indication of uterine problems. Approximately 5–10% of women with postmenopausal vaginal bleeding may have endometrial cancer.[6] Endometrial evaluation should be performed carefully in patients with postmenopausal bleeding. EAPs in postmenopausal bleeding women may lead to diagnostic difficulties. It is an unusual benign disease, accounting for only 1.3% of all endometrial polyps,[7] and may be mistaken for endometrial or cervical cancer. Histologically, EA is composed of endometrial-type glands surrounded by mature smooth muscle (the predominant component). When polypoid (EAP), it should be differentiated from APA, however, the latter has a more glandular component and less smooth muscle and does not show features of leiomyoma variants.[3] One retrospective case review revealed the EAP was often located at the uterine fundus, as in our case.[8] Similar to our case, Matsumoto et al[8] and Cheng et al[9] found some malignant changes such as endometrial and serous adenocarcinoma accompanied by APA.

The ultrasonographic pictures of EAPs are generally solid, well-circumscribed endometrial masses that range from 0.3 cm to 17 cm in diameter.[10] In one case series, the ultrasonographic features of the EAP constituted a solid mass with a cystic area, which was frequent and observed in 24 of 32 patients. Lee et al[10] also observed that APA, EAP, and endometrial hyperplasia are hard to distinguish using ultrasound. The EAP is a benign tumor, and submucosal adenomyoma should be carefully evaluated to distinguish APA, endometrial carcinoma, and endometrial sarcoma. In our case, the ultrasonographic scan revealed occupied lesions in the uterine cavity mimicking endometrial hyperplasia or cancer (Figure 1). Multiloculated cystic lesions were also noted. In postmenopausal women, thickened endometrium may arise due to malignant uterine changes.

The conventional MRI findings of EAPs comprise a well-defined polypoid mass protruding into the endometrial cavity, which is isointense relative to the myometrium, and contains high signal foci on T1- or T2-weighted images.[11] Our case revealed abnormal intrauterine lesions with a multiloculated cystic appearance and soft tissue components. In T2-weighted images, the picture (Figure 2) showed heterogeneous T2 prolongation, mimicking malignant imaging manifestation.[12]

Our case denied using tamoxifen, but had used hormone therapy for menopausal syndrome for 2 years, which was discontinued 3 years previously. However, the relationship between EAPs and hormones is not known. One case report found that gonadotropin-releasing hormone agonist treatment could decrease the size of EAPs.[13] The estrogen may be linked to formation of EAPs, explaining why EAPs rarely occur in postmenopausal women. The cause of adenomyoma in our case might be related to hormone therapy. Nevertheless, the relationship between adenomyoma and hormones requires further investigation.

The Pipelle endometrial biopsy was performed at the OPD and revealed only hematoma, which was poor in comparison with the imaging finding. Visser et al[14] observed that the amount of tissue obtained with outpatient endometrial sampling was insufficient for diagnosis of endometrial cancer in 30% of samples. According to their studies, atypical hyperplasia was noted in 13% of preoperative endometrial samples.[14] However, the final diagnosis of these cases following hysterectomy was endometrial cancer.[15] The Pipelle may have diagnostic pitfalls in EAPs such as in our case. One report revealed that the sensitivity of Pipelle and curettage was 93.8% and 97%, respectively, in patients with low-grade cancer, and 99.2% and 100%, respectively, in patients with high-grade cancer.[16] Pipelle may play a role in diagnostic preoperative endometrial samples, but some may overlook the malignant lesions. Therefore, hysteroscopy with transcervical resection was performed in this study to test the tissue and avoid unnecessary hysterectomy.

Hysteroscopy is very successful in the detection of endometrial lesion.[17,18] In postmenopausal bleeding, systemic evaluation of the endometrium by hysteroscopy raises the accuracy in the diagnosis of endometrial cancer.[19,20] In postmenopausal bleeding or endometrial thickening, hysteroscopy is a useful tool with high sensitivity and specificity.[21]

In conclusion, we presented one case with an EAP, which is a rare benign tumor of the uterus and difficult to distinguish from endometrial cancer using ultrasound or MRI. Diagnostic hysteroscopy is indicated if the result of tissue sampling by Pipelle does not correspond with the image findings.

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