A case of endometrial carcinoma arising in a 36-year-old woman with uterine atypical polypoid adenomyoma (APA)

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Summary

Atypical polypoid adenomyomas are tumors of low malignant potential. We present a case of endometrial carcinoma arising in a 36-year-old woman with atypical polypoid adenomyoma. The diagnosis and treatment of such a tumor is discussed through an English literature review.

Key words: Atypical polypoid adenomyoma (APA); Endometrial cancer; Differential diagnosis; Treatment; Prognosis.

Introduction

Endometrial polyps are lesions of the endometrial cavity which usually present with abnormal uterine bleeding. A less usual category of such lesions are atypical polypoid adenomyomas (APA). Longacre et al proposed that these lesions could be designated as low malignant potential tumors [1]. The differential diagnosis should be made from atypical endometrial hyperplasia, infiltrating carcinoma or malignant mixed mesodermal tumors.

Case Report

We present a case of a 36-year-old nulliparous obese woman with a history of menometrorrhagia for the previous three years. Pelvic examination was normal. The Papanicolaou smear was negative and transvaginal ultrasound showed a hyperechogenic lesion measuring 30 x 20 mm with poorly defined margins. The patient underwent dilatation and curettage which revealed an endometrioid endometrial cancer with grade 1 squamous differentiation. The patient underwent further investigation. Tumor markers (CA 19-9, CA15-3, CA125) were within normal ranges. MRI showed a hypointense endometrial lesion with hyperintense foci measuring 3.6 cm on T2-weighted magnetic resonance (MR) images and computed tomography (CT) revealed no enlarged lymph nodes. The patient underwent a total abdominal hysterectomy with bilateral salpingo-oophorectomy plus bilateral lymphadenectomy up to the level of the common iliac arteries. Cytologic analysis of free peritoneal fluid was negative. The histologic diagnosis was APA (Figure 1) with focal areas of invasive endometrioid endometrial carcinoma grade 1 (Figure 2) which invaded less than half of the endometrium (Stage Ia). No lymph node metastasis was found. The decision of the oncologists was follow-up with no need of further treatment. The patient was free of disease six months postoperatively.

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Discussion

APAs are usually found in premenopausal women. However, in the literature we were able to find some cases of APA in postmenopausal women [2-4]. Longacre *et al*. in 1996 reported that 96% of 55 patients with APA were premenopausal with a median age of 39 years. In the same study, 28/55 patients were nulliparous, 15/55 had a history of infertility and 13/55 were obese [1]. Moreover, it should be mentioned that APA has also been described in patients with Turner's syndrome [5]. For these reasons, hyperestrogenism could be proposed to be the pathogenetic mechanism. The question which arises is whether the lesion is related with endometrial cancer. Our case is one of the rare cases of the coexistence of endometrial cancer with APA [6-8]. It was found that some APAs exhibit MLH-1 promoter hypermethylation with focal lack of MLH-1 immunostaining, a molecular abnormality involved in the transition from complex atypical hyperplasia to endometrioid adenocarcinoma [9].

It should be noted that ultrasound characteristics of APAs (heterogeneous or homogeneous isoechogenic polypoid tumors with solid areas ± cystic areas and poorly defined margins) can not usually help in making the diagnosis preoperatively as the clinical suspicion should be increased to do so [10]. Furthermore, Kimura *et al*. suggested that even endometrial smears or biopsies might be inaccurate methods for the diagnosis of APA [11].

Mazur *et al*. reported the histologic characteristics of APAs in 1981. Microscopically APA is characterized by atypical and hyperplastic glands within stroma which are surrounded by smooth muscle [12, 13]. Cytologic atypia is characterized by enlarged nuclei with prominent nucleoli. The smooth muscle might show some mitotic activity (< 2 mitoses/10 hpf). Sometimes extensive squamous metaplasia with central necrosis may coexist, a fact which raises suspicions of carcinoma. However, carcinomas usually show greater cytologic atypia and more glandular crowding and architectural complexity. It should be noted that the stroma in APA and adenocarcinoma may show similar immunohistochemical markers such as smooth muscle actin, desmin and CD 34 [14].

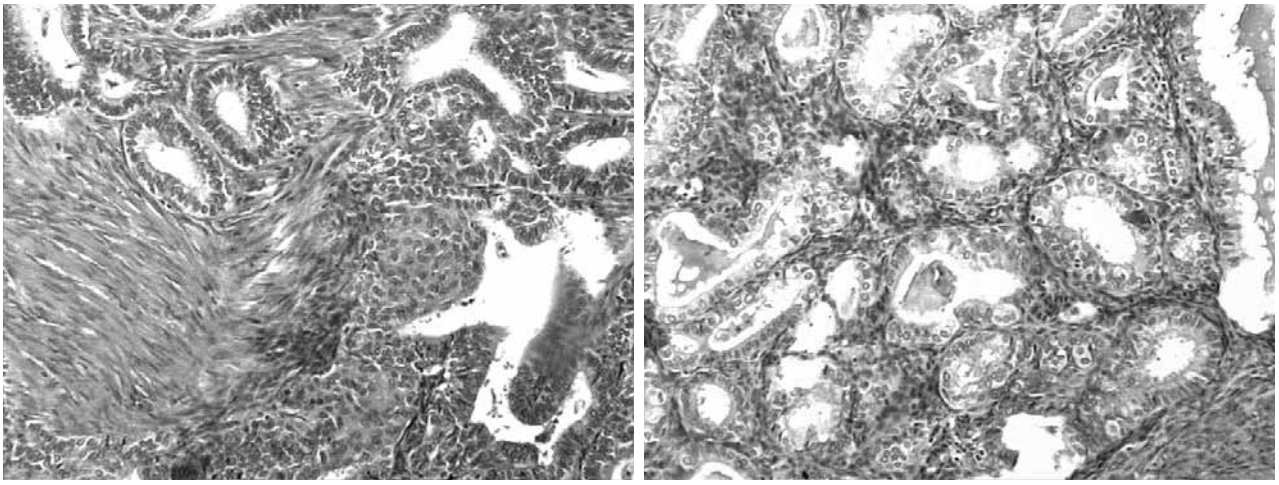

Figure 1. — Histological section of the uterine polypoid tumor showing smooth muscle fibers among endometrial glands (APA) (hematoxylin & eosin x 250).
Figure 2. — Histological section of APA showing the focal neoplastic component (hematoxylin & eosin x 150).

APA is a low malignant potential tumor which could be cured by curettage in order to permit the preservation of fertility. Furthermore, Vilos et al proposed hysteroscopic resection of such tumors as a fertility sparing option [15]. In a recent case report from Hong Kong, a live pregnancy is described in a woman with APA [16]. However, it should be mentioned that Longacre *et al*. in their retrospective study found a 45% recurrence rate in APAs treated conservatively. For this reason, close follow-up of these patients is proposed so that such recurrences can be found in early stages.

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