

Endometrioid adenocarcinoma arising in endometriosis foci six years after estrogen replacement therapy: a case report

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Summary

We present a case of a 53-year-old woman who developed an endometrioid adenocarcinoma six years after total abdominal hysterectomy (TAH) and bilateral salpingo-oophorectomy (BSO), who was on estrogenic-only hormone replacement therapy (HRT).

Key words: Endometrioid adenocarcinoma; Endometriosis foci; Estrogenic replacement therapy.

Case Report

We present a case of a 53-year-old woman, para 2, who attended her physician for abnormal vaginal bleeding of two months duration. She had had hypertension for ten years, controlled and unchanged.

She had been submitted to a TAH and BSO for leiomyomata six years before, and had been on estrogen-only HRT (estradiol, 1.5 mg twice a week).

During the pelvic exam an ulcerous-vegetating vaginal lesion was detected and a biopsy was taken. Afterwards, the patient was submitted to transvaginal ultrasonography and abdominal-pelvic computed tomography which demonstrated a tumour, appearing to have the bladder as the primary site and extending to the surrounding regions (both ureters and the rectosigmoid colon).

An anterior pelvic exenteration, anterior resection of the sigmoid colon, subtotal vaginal resection, a Bucker deviation and a protective appendectomy were carried out.

The final histological exam revealed an endometrioid adenocarcinoma, G1, affecting the vagina, bladder and rectum; two pelvic lymph nodes were positive.

The patient underwent six cycles of chemotherapy with cisplatin (50 mg/m²) and doxorubicin (60 mg/m²).

The patient is currently alive without evidence of disease six months after the diagnosis.

Discussion

The patient has been submitted to TAH-BSO six years before for leiomyomata and no endometriosis foci were reported in the histological exam. Thus, she was placed on estrogen-only HRT.

After the histological results of the last surgery were known, the pathologist re-examined the histological findings of the TAH-BSO, which demonstrated that there was no endometrioid adenocarcinoma. There were however pelvic endometrioid foci (especially in the fallopian tubes), which had not been reported before.

Considering these results, the best hypothesis was that the endometrioid adenocarcinoma seen presently had evolved from pelvic endometrioid foci, stimulated by the estrogen-only HRT.

Favouring this hypothesis was: the fact that the tumour infiltrated by an extrinsic way the rectum and bladder; the endometrioid histological pattern; the existence of endometriosis foci near the neoplasia and the immunohistochemistry results (CK 7 positive, CK 20 negative and estrogen receptor positive).

Since 1940 some cases linking estrogen therapy with endometrial carcinoma have been reported [1].

There is also some evidence that endometriosis behaves in a malignant manner: it can be metastatic and connect to other tissues, invading and harming them [2].

In the literature, a 0.3 to 1% incidence of malignant transformation of endometriosis has been reported [3-5]. Such malignant transformation occurs mostly in the ovary but 25% of the cases occur in extragonadal endometriosis, most commonly in the rectovaginal septum [3, 5, 6]. We also know that the malignant transformation of endometriosis can lead to the appearance of any histological type but that endometrioid adenocarcinoma is the most encountered [7].

After TAH-BSO the malignant transformation of endometriosis can occur either by secondary implantation of endometriosis or by extra-genital endometriosis not reported [3].

Sampson in 1925 defined three diagnostic criteria for the malignant transformation of endometriosis: proximity of benign endometriosis with the malignant tumour, no other primary tumour identified and histology demonstrating a primitive endometrial tumour [7, 8].

It seems logical to presume that estrogen-only HRT leads to endometrial proliferation and by stimulating the cellular turnover, genetic errors become more probable leading to the appearance of abnormal cells, that once stimulated lead to the appearance of adenocarcinoma. As endometrioid foci behave like endometrial tissue, it is likely that this theory applies also to them.

Revised manuscript accepted for publication October 21, 2003

Conclusion

Although estrogen-only HRT is usually given to women submitted to TAH-BSO, we should not forget the initial surgical indications or the preoperative findings [3, 6, 7].

Thus, the recommendation is to give combined HRT (estrogens and progestins) to women with a history of endometriosis.

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