Case Reports

Malignant transformation of persistent endometriosis after hysterectomy

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ABSTRACT

من الحالات النادره تحول داء البطانة الرحمية المنتشر خارج الرحم بالحوض بعد إزالته جراحياً إلى ورم خبيث، لكن هناك عدة حالات سجلت ونشرت في المجلات العلمية المحكمة لهذه الحالة. استعمال المريض لهرمون الاستروجين التعويضي عامل مشترك بين هذه الحالات. سوف نستعرض حالة امرأة عمرها 53 سنة كانت مشخصه بداء البطانة الرحمية وخضعت للعلاج الجدري لهذه الحالة وذلك بازالة الرحم والمبايض. بعد 9 سنوات من ذلك، عانت المريضة من ورم خبيث في أسفل البطن وتم إزالته جراحياً، وأثبتت الدراسة المجهرية أنه من أصل البطانة الرحمية. كما سوف نناقش إماكنية تحول البطانة إلى ورم خبيث وكذلك العوامل المؤثره على العملية مع مراجعة سريعة حول الموضوع.

The malignant transformation of persistent endometriotic implants into endometrioid adenocarcinoma is rare, especially after remote hysterectomy and salpingo-oophorectomy (TAH-BSO), and there are few cases reported in the English language literature. Patients receiving estrogen replacement therapy are common among the reported cases. We present a case that demonstrates the possibility of malignant transformation in a 53-year-old female, known case of endometriosis, who underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy with no evidence of malignancy in the final pathology report. After 9 years, she presented with lower abdominal mass, and histopathological studies confirmed the diagnosis of well-differentiated endometrioid adenocarcinoma. The possibility of malignant transformation and possible risk factors are discussed with a brief literature review.

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Malignant transformation of persistent endometriotic implants into endometrioid adenocarcinoma is rare, especially after a remote total hysterectomy and salpingo-oophorectomy (TAH-BSO), and there are few cases reported in the English language literature.[1] Patients receiving estrogen replacement therapy are common among the reported cases.[2] We present a case that demonstrates the possibility of malignant transformation, a 53-year-old female, and a known case of endometriosis, who underwent TAH-BSO with no evidence of malignancy in the final pathology report. After 9 years, she presented with lower abdominal mass, and histopathological studies confirmed the diagnosis of well-differentiated endometrioid adenocarcinoma. The possibilities of malignant transformation and possible risk factors are discussed here with a brief literature review.

Case Report. The patient was a 53-year-old female, para gravida 2 with no known medical illness. She presented with lower abdominal pain and constipation for 2 months. There was no per vaginal, or rectal bleeding. The systemic review was unremarkable. Her surgical history was a known case of endometriosis with severe intra-abdominal adhesions (diagnosed by diagnostic laparoscopy). Thereafter, she underwent TAH-BSO 9 years ago. She was asymptomatic until

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this presentation. Her family history was negative for malignancy.

On examination, she was conscious and vitally stable. The cardiac and respiratory examination was unremarkable. Her abdomen was soft, lax, non-tender with a suprapubic mass of approximately 15x10 cm, firm in consistency and not fluctuating, and with a negative cough impulse. She had a full preoperative assessment made. Laboratory investigations, including a complete blood count and coagulation profile, were all within the normal range. The cancer antigen 125 (CA-125) was 86.7 UI/ml at presentation. The magnetic resonance imaging (MRI) with intravenous gadolinium injection showed a well capsulated large pelvic-abdominal mass 16.7x10x14 cm in size (**Figure 1**). The mass showed enhancing internal septation with a solid component in its right anterolateral wall. Enhancing the solid component, it had inner lobulation, papillary projections, and it was iso-intense to the muscles. There was no clear peri-lesion infiltration other than a small focal area close to the right external vessels. The rectum, sigmoid, and urinary bladder were compressed and displaced postero-laterally with no obvious focal infiltration. There was no peritoneal thickening or enhancement. The pelvic side wall was not involved. There was no lymphadenopathy and no free fluid. Our clinical suspicion was of a lower gastrointestinal mass, or a lower genitourinary-related mass; however, malignant neoplasm pathology was at the top of the list.

She underwent laparotomy and pelvic-abdominal mass excision following approval from the tumor board. A complete resection was carried out without any injury to the bowel, ureter, or pelvic organs. The mass was cystic in nature, containing approximately 2 liters of thick, coffee-colored fluid. A frozen section was sent during the operation that showed the cystic wall, which is likely to be endometriosis. The postoperative patient had an uneventful recovery without complications. Histological examinations of the specimens showed moderately differentiated endometrioid adenocarcinoma of highly atypical cells forming solid sheets and glands. The tumor cells had pleomorphic nuclei and high a nucleus/cytoplasm ratio. In addition, there are multiple areas of endometriosis. Immunohistochemistry showed estrogen receptors+, progesterone receptor++, cytokeratin 7++, and CK20-. She continued the follow up at the gynecology clinic, and started chemotherapy. The plan was to receive 6 cycles of chemotherapy (carboplatin) every 3 weeks followed by clinical follow up. After the 6 cycles of chemotherapy, she was symptomatic free, and her CA-125 was normalized.

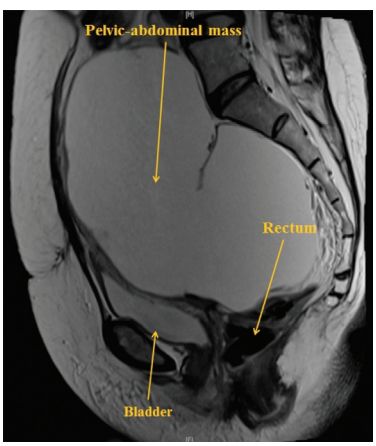

Figure 1 - An image showing a sagittal view of MRI T2 high signal intensity of the mass in the pelvis. The mass was compressing the rectum and urinary bladder.

Discussion. Endometriosis is a common disease in which functioning endometrial tissue (stroma and glands) is present outside the uterine cavity, or myometrium. The reported incidence is between 7 and 10%.[3,4] Pelvic organs are the most common targets for endometriosis (including the ovaries, pelvic cul-de-sac, and broad ligament, and so forth). The most commonly involved organ is the ovary. In addition, it has been described in numerous other locations in the pelvis and abdomen.[5] Malignant transformation of endometriosis is an uncommon event. Atypia (intraepithelial neoplasia) in endometriosis is seen in 2% of cases without a neoplasm,[6] and the estimated risk of cancer arising from preexisting endometriosis is approximately 0.7-1.0%.[7] Clear cell and endometrioid carcinoma are the most common histopathological types of cancer reported in women with ovarian endometriosis. Moreover, clear cell adenocarcinoma and adenosarcomas are the 2 most common malignancies arising in extra-ovarian endometriosis.[8] There is another theory that suggests that there is an increased risk of some cancers with this situation particularly in the ovary, non-Hodgkin's lymphoma, and breast cancer.[9]

There are reported cases in the literature describing the malignant transformation of endometriosis even after definitive surgery (TAH-BSO).[2] Sampson's criteria was used to confirm that the tumor arises from an endometriosis deposit; the histopathology of this mass should be of endometrial origin, endometriosis, and malignant tissue are found in close proximity, and no other localization of primary tumor.[10] Sampson's criteria fits our case except there is no proximity of endometrial tissue, since she had remote TAH-BSO. The risk factors for this process include prolonged

exposure to unopposed estrogen and obesity, even after a definitive surgery. Because of this, post-hysterectomy combining estrogen and progesterone treatment has been recommended in patients with suspected residual endometriosis.[2-8] However, our patient did not receive any hormonal replacement therapy, nor was she overweight.

In conclusion, the recurrence of endometriosis after TAH-BSO is possible, as well as malignant transformation into cancer. Unopposed estrogen use may increase the risk of this malignant transformation. It requires a multidisciplinary approach to manage this type of case.

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