Case Report

Cystic Uterine Endosalpingiosis in a Patient with Carcinoma Endometrium

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Abstract

Endosalpingiosis is a benign condition characterized by the presence of tubal-type epithelial cells outside the Fallopian tube. It may rarely involve the uterus and present as a cystic or tumor-like mass. We report an unusual case of cystic uterine endosalpingiosis in a postmenopausal female with carcinoma endometrium. Preoperative and intraoperative diagnosis of this condition is challenging. Awareness about this condition in clinicians may help in preventing misdiagnosis and overtreatment.

Keywords: *Carcinoma endometrium, cystic uterine endosalpingiosis, müllerianosis*

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Introduction

Endosalpingiosis is a condition characterized by the presence of glands lined by tubal-type epithelial cells outside the Fallopian tube. It usually involves peritoneum and omentum but rarely may involve lymph nodes, bowel, and urinary system.[1,2] It may also rarely involve the uterus presenting as a cystic or tumor-like mass.[3] This benign condition is usually asymptomatic. We report a case of cystic uterine endosalpingiosis which was incidentally detected in a patient with carcinoma endometrium.

Case Report

A 53-year-old woman, gravida 2, para 2, was referred to our hospital with complaints of postmenopausal bleeding. She had attained menopause 4 years back and had an episode of vaginal bleeding 1 week back. She had no complaints of pelvic pain. Pelvic examination revealed a bulky uterus and no adnexal mass. Transvaginal sonography was done which showed minimal endometrial collection with an endometrial thickness of 2 cm. It also showed multiple cysts in the adnexal region. The patient underwent hysteroscopic-guided biopsy which revealed Grade 1 endometrioid carcinoma of the endometrium. The patient was planned for the surgery and a pelvic contrast-enhanced computed tomography was done. It showed a 3-cm growth in the endometrium without any myometrial invasion and multiple cysts in the adnexal region and was reported as ovarian cysts [Figure 1]. CA 125 levels of the patient were in normal limits.

Intraoperatively, the uterine surface was found to be studded with multiple cysts filled with clear fluid [Figure 2]. Bilateral Fallopian tubes and ovaries were normal in appearance. Extrafascial hysterectomy, bilateral salpingo-oophorectomy, and pelvic lymphadenectomy were done. On histopathological examination, the serosal surface of the uterus showed many multiloculated cysts, with cyst wall thickness measuring 0.1–0.2 cm. The endometrial cavity showed a tumor which was gray white involving <50% of the myometrium. The cervix, parametrium, bilateral tubes, and bilateral ovaries were unremarkable.

Microscopically, the endometrial cavity showed a tumor suggesting Grade 1 endometrioid carcinoma with <50% myometrial invasion. Uterine serosal cysts were lined by ciliated and nonciliated cuboidal-to-columnar epithelium with underlying smooth muscle [Figure 3]. These cells showed bland nuclear features, no mitotic figures. The

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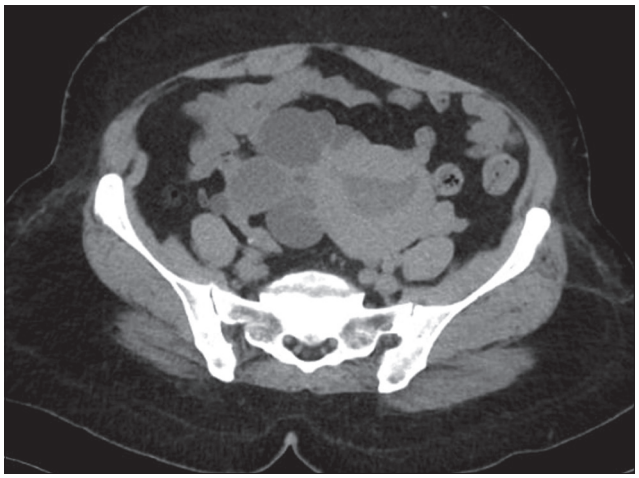

Figure 1: Contrast-enhanced computed tomography of the pelvis showing the uterus with endometrial collection and multiple adnexal cysts

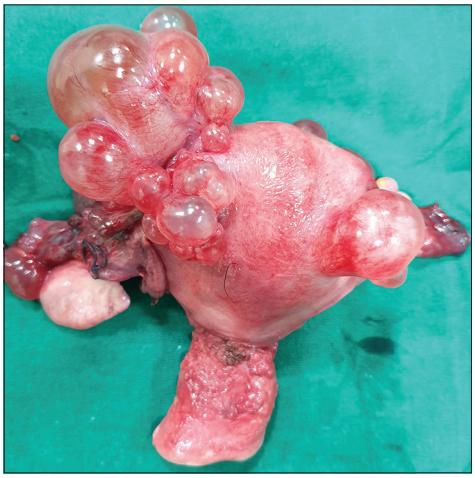

Figure 2: Multiloculated clear cysts arising from the uterine serosa

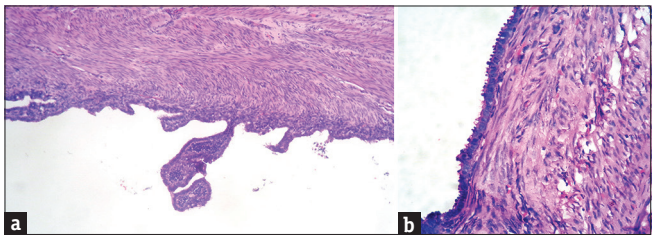

Figure 3: (a) ×10 (section shows cystic lesion with lining simple columnar cells [secretory epithelium]) (b) ×40 (high-power view showing simple columnar cells [secretory cells and ciliated epithelial cells])

epithelium resembled normal tubal epithelium with focal areas showing papillary infolding, suggesting endosalpingiosis. A diagnosis of Stage 1a carcinoma endometrium with uterine cystic endosalpingiosis was made. The patient did not receive any adjuvant treatment and is currently on follow-up in our cancer clinic every 3 months. No additional treatment is planned for the patient.

Discussion

Endosalpingiosis along with endometriosis and endocervicosis, are sometimes referred to as "Müllerianosis."[4] When the coelomic epithelium lining the peritoneal cavity undergoes differentiation into glands with histologic features of tubal epithelium, it is termed as endosalpingiosis. It is usually found in women of reproductive age but may occur in postmenopausal women as well. In a recent study, 40% of endosalpingiosis cases occurred in postmenopausal women.[5] They also reported concurrent endometriosis in 34.5% of all endosalpingiosis patients.

The pathogenesis of this condition is not clearly known but may be similar to endometriosis. Ectopic transport of cells has been suggested as one of the theories.[3] Previous abdominal surgeries, especially involving the Fallopian tubes or ovaries, have been reported as risk factors.[6] In a study involving 838 women with endosalpingiosis, 78.3% of women had a history of prior abdominal surgery.[7] Although most of the patients are asymptomatic, some may present with complaints such as pain abdomen, menorrhagia, or urinary symptoms. It has not been found to be associated with infertility or chronic pelvic pain.[5]

Most of the patients with uterine endosalpingiosis have been treated with hysterectomy. In a recent study, it was reported that up to 75% of patients with uterine endosalpingiosis underwent a hysterectomy and 62.5% underwent bilateral salpingo-oophorectomy.[3] In a young symptomatic patient, mass excision can be offered as an alternative to hysterectomy.

Cystic endosalpingiosis involving uterus or ovary may be mistaken as malignancy during surgery because of its gross appearance.[8] CA 125 level has been reported to be normal in most of these patients.[3,8,9] Although it is a benign condition, some studies have reported malignant transformation in these patients.[10] Further, some studies have reported a higher risk of gynecological malignancies in patients with endosalpingiosis.[5,7] Our patient had endometrioid adenocarcinoma of the endometrium and was also found to have cystic endosalpingiosis of uterine serosa. This cystic endosalpingiosis was an incidental finding and not directly related to the underlying malignancy. Preoperative imaging in our patient had diagnosed multiple ovarian cysts. Intraoperatively, there were multiple cysts covering the uterine surface. It may be possible sometimes to misdiagnose these lesions as tumor deposits on uterine serosa. Although this condition is rarely encountered in routine clinician practice and presented here mainly for its unique nature, awareness about this rare condition of uterus may help in preventing misdiagnosis and overtreatment.

Conclusion

Uterine cystic endosalpingiosis is a rare condition which may be misdiagnosed as ovarian cyst preoperatively and as a malignant tumor deposit intraoperatively. If symptomatic, patients can be offered conservative surgery provided there is no underlying malignancy. Awareness about this rare condition in clinicians may help in preventing misdiagnosis and overtreatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

References

1. Zinsser KR, Wheeler JE. Endosalpingiosis in the omentum: A study of autopsy and surgical material. Am J Surg Pathol 1982;6:109-17.
2. Habiba M, Brosens I, Benagiano G. Müllerianosis, endocervicosis, and endosalpingiosis of the urinary tract: A literature review. Reprod Sci 2018;25:1607-18.
3. Yang M, Li Y, Chen M, Chen J, Kung FT. Uterine endosalpingiosis: Case report and review of the literature. Taiwan J Obstet Gynecol 2019;58:324-7.
4. Batt RE, Yeh J. Müllerianosis: Four developmental (embryonic) Mullerian diseases. Reprod Sci 2013;20:1030-7.
5. Prentice L, Stewart A, Mohiuddin S, Johnson NP. What is endosalpingiosis? Fertil Steril 2012;98:942-7.
6. Ong NC, Maher PJ, Pyman JM, Readman E, Gordon S. Endosalpingiosis, an unrecognized condition: Report and literature review. Gynecol Surg 2004;1:11-4.
7. Esselen KM, Terry KL, Samuel A, Elias KM, Davis M, Welch WR, *et al*. Endosalpingiosis: More than just an incidental finding at the time of gynecologic surgery? Gynecol Oncol 2016;142:255-60.
8. Singh N, Murali S, Zangmo R. Florid cystic endosalpingiosis, masquerading as malignancy in a young patient: A brief review. BMJ Case Rep 2014:bcr2013201645. doi: 10.1136/bcr-2013-201645.
9. Zapardiel I, Tobias-Gonzalez P, de Santiago J. Endosalpingiosis mimicking recurrent ovarian carcinoma. Taiwan J Obstet Gynecol 2012;51:660-2.
10. McCoubrey A, Houghton O, McCallion K, McCluggage WG. Serous adenocarcinoma of the sigmoid mesentery arising in cystic endosalpingiosis. J Clin Pathol 2005;58:1221-3.