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SILENT PATHOLOGY ENDOSALPINGIOSIS: CASE SERIES

SESSİZ PATOLOJİ ENDOSALPİNGİOSİS: VAKA SERİSİ

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ABSTRACT

Objective: The aim of this study was to evaluate the clinical data on endosalpingiosis, which is only pathologically recognized, and which may accompany malignancies and endometriosis.

Material and Method: We collected the data of patients who were pathologically diagnosed with endosalpingiosis between May 2017 and January 2023 in a tertiary health institution. The patient's age, gravida, clinical complaints, imaging findings, presence of known risk factors for endosalpingiosis, the operation performed, and the location of the endosalpingiosis focus were recorded. The postoperative statuses of the patients were provided by hospital records and telephone interviews with the patients.

Result: The data of six female patients between 34 and 62 years of age were analyzed. Two of the patients were in menopause, five had a history of previous abdominal surgery, one had endometriosis, and one patient had endosalpingiosis accompanying high-grade serose ovarian carcinoma.

Conclusion: Considering the increase in synchronous malignancies, we believe that endosalpingiosis should be reported by pathology, especially in cases of endometriosis and those with a history of previous surgery, and the clinician should inform the patient of the presence of this diagnosis.

Keywords: Endosalpingiosis, endometriosis, mullerianosis, ovarian cancer

ÖZET

Amaç: Bu çalışmanın amacı sadece patolojik olarak tanınan ve malignitelere ve endometriosise eşlik edebilen endosalpingiosis ile ilgili klinik verileri değerlendirmektir.

Gereç ve Yöntem: Tersiyer sağlık kuruluşunda Mayıs 2017-Ocak 2023 arası patolojik olarak endosalpingiosis tanısı konmuş hastaların verilerini topladık. Hastaların yaş, gravida, klinik şikayet, görüntüleme yöntemi bulguları, endosalpingiosis için bilinen risk faktörlerinin varlığı, yapılan operasyon, endosalpingiosis odağının yeri kaydedildi. Hastaların operasyon sonrası durumları hastane kayıtları ve hastalar ile yapılan telefon görüşmeleri ile sağlandı.

Bulgular: Altı kadın hastaya ait veriler incelendi. Hastalar 34-62 yaş arasındaydı. İki hasta menapozdaydı. Beş hastada geçirilmiş batın ameliyatı öyküsü vardı. Bir hastada endometriosis de vardı. İki hastada geçirilmiş appendektomi, iki hastada sca öyküsü, iki hastada RİA vardı, bir hastada ektopik gebelik nedeniyle salpenjektomi yapılmıştı. Bir hasta high grade seröz over kanseri eşlik eden endosalpingiosis vardı.

Sonuç: Senkron malignitelerdeki artış göz önüne alınarak özellikle endometriosis vakalarında ve geçirilmiş cerrahi öyküsü olanlarda endosalpingiosisin patoloji tarafından rapor edilmesi gerektiğine ve klinisyenin bu tanı varlığında hastayı bilgilendirmesi gerektiğine inanıyoruz.

Anahtar Kelimeler: Endosalpingiosis, endometriosis, mullerianosis, over kanseri

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INTRODUCTION

Endosalpingiosis, first described by Sampson in 1930, is the presence of columnar ciliary glandular epithelia similar to the tubal epithelium outside the tuba. It is a benign and rare entity that is most commonly seen in the pelvic peritoneum covering the uterus, ovary, and tubule, and less commonly in the omentum, lymph nodes, cervix, etc. (5). It can be seen in many different guises, such as small, round, transparent vesicles, light-colored or red smooth-surfaced peritoneal polyps, or opaque, fimbria-like buds (1). In addition, it can be seen as a single cyst with a flat surface, as sarcoma-like red lesions, or as intrauterine cysts (2).

The frequency of endosalpingiosis is not clearly known due to it not being reported by pathology and the lack of a known method of diagnosis, but in one study, the frequency was found to be 7-12% in gynecological surgeries performed for any reason (3).

The coexistence of endosalpingiosis, endocervicosis and endometriosis is defined as mullerianosis (2). Endosalpingiosis is most commonly seen with endometriosis (40%). It can also be seen with some low-grade and borderline malignancies. In cases not associated with endometriosis, endosalpingiosis and synchronous ovary cancer are seen at a younger age (mean 39 years) (4).

The pathogenesis of endosalpingiosis is not clear. However, multifocal metaplasia from the mesothelium, and detection in the lymph node in male patients with prostatic carcinoma supports mullerian metaplasia. Another theory is tubal mucosal implantation. The fact that endosalpingiosis is more common in patients with a previous surgical history supports this theory. Three quarters of those with endosalpingiosis have a history of surgery (6).

Complaints associated with endosalpingiosis may be related to the accompanying disease. In addition, symptoms related to the affected organ such as menorrhagia, abnormal uterine bleeding, abdominal distension, difficulty urinating, and polyuria may be observed. In a gynecological evaluation, this condition can be confused with leiomyoma, adenomyoma, adenocarcinoma, uterine sarcoma, peritoneal tumors, and ovarian cancer (2).

Since biomarkers similar to those for a tubal tumor are secreted in endosalpingiosis, it has been named in the literature as a low-grade or borderline tubal tumor. Studies have shown that ovarian and uterine cancers increase, especially clear-cell carcinomas and mucinous tumors, depending on the capacities of cells with metaplasia in endosalpingiosis cases (3). Nevertheless, endosalpingiosis is a non-neoplastic pathology, and radical oncological surgery is not recommended for the treatment of it.

Our aim in this study is to draw attention to the fact that endosalpingiosis is a pathology that can develop in the genital and other organs and tissues and may accompany malignancy and endometriosis.

MATERIAL and METHODS

This retrospective study was started after the approval of the ethics committee (Date: 13.04.2023, No: 387), and verbal and written consent was obtained from the patients included in the study. The data of six patients who were operated on due to gynecological complaints at the Gynecology and Obstetrics Clinic of the tertiary center between May 2017 and January 2023 and were diagnosed with endosalpingiosis in the pathological examination were collected and evaluated through hospital records and interviews with the patients themselves.

The age, gravida, menopausal status, previous surgery history, previous malignancy history, admission complaint, remarkable laboratory value, ultrasonography and magnetic resonance imaging and Doppler findings (if performed), surgery performed, pathological diagnosis accompanying gynecological pathology, and postoperative conditions of the patients were recorded.

RESULTS

The data of six female patients diagnosed with endosalpingiosis over five years were analyzed. These patients were aged between 34 and 62 years, two of whom were in menopause.

Endosalpingiosis was found in the resected sigmoid colon serosa in one patient, in the ovary in two patients, in the paraovarian area in one patient, in the pelvic lymph node in another patient, and in one patient, endosalpingiosis was in the perimetrium.

All the patients had had a previous pregnancy.

Five patients had a history of previous abdominal surgery. Two patients had previous appendectomy, two had a history of sectio caesarea abdominalis (SCA), two had intrauterine devices (IUDs), and one had a salpingectomy due to an ectopic pregnancy. One patient had endosalpingiosis accompanying high-grade serose ovarian cancer.

One patient also had synchronous endometriosis. There were paratubal cysts in three patients.

There was no additional medicine used by the patients.

Two of the patients had abnormal uterine bleeding and one had postmenopausal bleeding.

Preoperative peritoneal nodularity was suspected in one of the patients, and in the other patients it was found incidentally.

Total abdominal hysterectomy and bilateral salpingo-oopherectomy were performed on five patients. One patient was diagnosed during sigmoid colon surgery.

Information about the patients, their previous surgery history, and pathological features are shown in Table 1.

Except for the appearance of peritoneal nodules in one patient, there was no feature that aroused suspicion of endosalpingiosis in the USG, Doppler and MRI images of the patients.

All of the patients are alive, including the ovarian cancer case.

DISCUSSION

The median age at diagnosis of endosalpingiosis is 50-52 years (10). The patients in the study were on average 47 years old.

There is more than one theory for the etiology. While the metaplasia theory is valid in some patients as it can accompany malignancy and borderline tumors, the explanation in women who have undergone tubal surgery, women with abnormal uterine bleeding and women using IUDs is the tubal cell implantation theory.

Endosalpingiosis has no known macroscopic or imaging features, and often millimetric structures are seen as vesicular effects (1). Diagnosis is made only by microscopic examination. In the study, there was no remarkable abnormality except for paratubal cysts in three patients and a purpura-like lesion on the tubal surface in one patient. Although ultrasonography was performed on all patients and MRI imaging was performed on some patients, endosalpingiosis was diagnosed microscopically in only one patient with suspected peritoneal nodularity on MRI.

Although endosalpingiosis is a benign and silent pathology, it is important in terms of the possibility of accompanying pelvic serous neoplasms (such as lesions of low malignant potential, or low-grade pelvic serous carcinoma). Some data suggest that endosalpingiosis is more common in women with BRCA mutations. In a study that found endosalpingiosis in 86% of BRCA carriers who underwent risk-reducing salpingo-oophorectomy, endosalpingiosis was reported in 56% of women at low risk of ovarian cancer who underwent salpingo-oophorectomy for other indications (7). Our 39 years old patient had high-grade serose ovarian carcinoma (HGSCa) as well as endometriosis and endosalpingiosis. It is seen in the literature that there is a relationship between borderline ovarian tumor (BOT) and endosalpingiosis. However, endosalpingiosis was not detected in our BOT patients in that study (8). In the literature, an increase in malignancies accompanying endosalpingiosis in premenopausal women is mentioned. In our study, we found endosalpingiosis as well as endometriosis and HGSCa in one young premenopausal patient, which may make it meaningful to perform frozen lesion next to a pathology that is thought to be of no clinical importance.

We believe that performing a biopsy for abnormal appearances, such as atypical cystic, nodular polypoid appearances, or increased vascularity in a patient who has been operated on for any reason, may help determine the malignancy risk at that time or later. The prevalence of serous borderline, invasive mucinous, and clear cell histological subtypes is increased in patients with ovarian cancer and endosalpingiosis.

Table 1: Demographic clinical characteristics of the patients

Number	Age (years)	Gravida (n)	Operation-indication	Post-surgery time	Risk factors	Place
1	34	4	Sigmoid colon resection-diverticule perforation	4 years	Appendectomy-intrauterine device	On the colon serosa
2	49	4	TAH+BSO-myoma uteri and abnormal uterine bleeding	6 years	Umbilical hernia operation-endometrial probe curettage	On the ovary
3	53	1	TAH+BSO-myoma and postmenopausal bleeding	5 years	Ectopic pregnancy (salpingectomy)	Bilateral ovaries
4	39	1	Debulking-ovarian Ca and abnormal uterine bleeding	2 years	Appendectomy-Intrauterine device	On the pelvic lymph node
5	62	6	TAH+BSO-postmenopausal bleeding and endometrial hyperplasia	5 years	None	On the ovary
6	45	3	TAH+BSO-myoma	6 years	Bilateral tubal ligation-SCA	Perimetrium

TAH: total abdominal hysterectomy BSO: Bilateral salpingo-oophorectomy, y: SCA: Sectio caesarea abdominalis

Transplantation of the tubal mucosa to the peritoneum in patients with a history of previous surgery or excessive proliferation and healing after tubal surgery is thought to be effective (6). Five of the six patients included in this study had a history of abdominal surgery.

We encountered endosalpingiosis, defined as mullerianosis, with additional endometriosis in one of our patients. Mullerianosis supports an embryological etiology (9). Endometriosis occurs simultaneously in more than one-third of patients with endosalpingiosis.

Although there is information about the regression of endosalpingiosis when the ovaries are removed, two of the patients in this study were in menopause. This supports a hormone-independent etiology (6).

The small number of cases in this clinical study prevents us from defining the clinical presentation of complaints of endosalpingiosis and classifying the accompanying pathologies. However, endosalpingiosis, which is considered to be clinically insignificant, cannot easily be diagnosed due to unperformed biopsies and is recognized only by examining a large area in organ removal surgeries. Our study is, therefore, retrospective and still valuable.

In many cases operated for gynecological reasons, it is not easy to decide whether to perform cystectomy, oophorectomy or salpingo-oophorectomy. Especially if some patients have peritoneal or tissue lesions suggestive of endosalpingiosis, it may be easier to decide by considering the endosalpingiosis malignancy relationship (for example, salpingectomy instead of tubal ligation, or oophorectomy instead of cystectomy).

In this study, we wanted to draw attention to the importance of accurate reporting of endosalpingiosis by pathologists, especially in cases of endometriosis and those with a previous surgical history, considering the increase in synchronous malignancies.

Ethics Committee Approval: This study was approved by Medipol University Non-Interventional Clinical Researches Ethics Committee (Date: 13.04.2023, No: 387).

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