

Research Letter

Endosalpingiosis mimicking recurrent ovarian carcinoma

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Endosalpingiosis is defined as the presence of glandular nests covered by tubal epithelium in the peritoneum or on the serous surface of the uterus and ovaries [1]. It is a rare entity, which normally appears in women of reproductive age, with an average age of presentation between 30 and 50 years [2–4]. It often appears alone, but it is not unusual to find endosalpingiosis associated with endometriosis or endocervicosis [5]. It has been theorized that it may be involved in the pathogenesis of ovarian tumors, although not enough evidence has been found to confirm this hypothesis [6].

The diagnosis of endosalpingiosis is made histologically. It usually consists of a layer of simple columnar epithelium formed by ciliated cells, non-ciliated secretory cells and peg cells. In addition, it can form papillae, show psammoma bodies and be surrounded by chronic inflammatory cells [3]. Sometimes it can mimic neoplasms and peritoneal tuberculosis on a macroscopic level, making a difficult differential diagnosis [7,8].

We report the case of a postmenopausal woman with a past history of early-stage ovarian cancer who presented with an extended endosalpingiosis that resembled malignant peritoneal recurrence.

A 68-year-old woman presented at our outpatient clinic complaining of abdominal bloating and nausea during the past 3 months. She had undergone a simple hysterectomy 31 years previously for bleeding, and she had received a bilateral salpingo-oophorectomy 9 years ago for an International Federation of Obstetricians and Gynaecologists (FIGO) stage IA ovarian cancer with no adjuvant chemotherapy.

On physical examination, there was a palpable mass in the midpelvic area and the vagina appeared normal. An abdominopelvic computed tomography scan revealed a bi-lobed 8-cm mass with cystic and perhaps solid components in the pelvis draped over the rectum and just inferior to the tortuous sigmoid colon. This might have arisen from the vaginal cuff

and raised suspicion for recurrent ovarian malignancy. No abdomen or pelvis adenopathy was found and there was no evidence of free fluid. Serum tumor markers were negative.

The patient underwent laparoscopic surgery. Extensive involvement of the pelvic peritoneum, anterior abdominal wall peritoneum, serosa of the sigmoid (Fig. 1) and vaginal cuff of cystic implants (resembling a pelvic mass in the pelvis) was found. The cystic implants were visually consistent with recurrent borderline/invasive ovarian carcinoma, but on frozen section they were interpreted as probably endosalpingiosis. Awaiting the final pathology report to make the differential diagnosis with low malignant potential tumor and considering the patient's cancer history, she also underwent upper vaginectomy, complete pelvic peritonectomy and excision of the bladder peritoneum, lower anterior abdominal wall peritoneum and cystic lesions of the sigmoid. Infracolic omentectomy was also performed. The patient had an uneventful postoperative course but she was not able to void properly until day 2. She was discharged on postoperative day 3. The final pathological report informed of extensive fibroadipose tissue linked to several cystic structures lined by ciliated epithelium and consistent with benign serous cysts of the peritoneum and endosalpingiosis. At 1 year of follow-up, the patient remains asymptomatic.

The term endosalpingiosis was first used in 1930 by Sampson, who observed the presence of tubal epithelium in the scars of previous salpingectomies [9]. Sampson considered that the origin of endosalpingiosis lay in the invasive nature of the tubal mucosa, essentially being the presence of a previous salpingectomy, salpingitis, or chronic pelvic inflammatory process.

The pathogenesis of endosalpingiosis is still controversial, and two main theories have been proposed. For some authors, the origin of endosalpingiosis is considered to be the peritoneal implantation of sloughed tubal epithelium and more rarely hematogenous or lymphatic spread [3,9]. The other theory, which is more commonly accepted nowadays, suggests that endosalpingiosis is due to a metaplasia of mesothelial peritoneal cells [2,10,11]. These cells would be part of the

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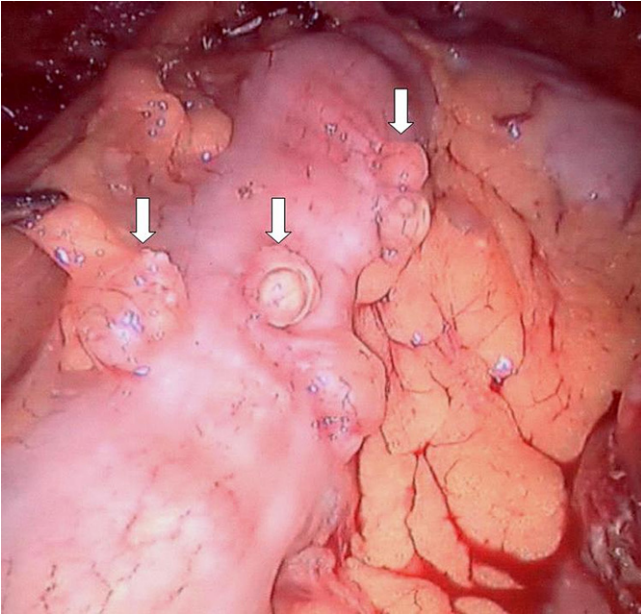


Fig. 1. Suspicious lesions (white arrows) on the sigmoid colon.

secondary Müllerian system (the pelvic peritoneum), which is different from the primary Müllerian system that is responsible during embryogenesis for the formation of the fallopian tubes, the uterus, the cervix, and the upper part of the vagina [12]. This last theory is supported by the association described between endosalpingiosis and ovarian serous low-grade tumors [10], as the presence of endosalpingiosis is observed in 33–71% of cases of ovarian serous low-grade tumors [13,14]. Due to these findings, it has been proposed that endosalpingiosis may be a precursor lesion, as even malignant processes such as serous adenocarcinoma have been described in previous endosalpingiosis [15]. In the case reported, the patient's age and her previous history of ovarian cancer made us suspect a relapse of the disease. The disease appears most commonly in premenopausal women, although here we report a case in a postmenopausal woman.

Regarding its clinical presentation, endosalpingiosis does not have specific symptoms, and in most cases it is an asymptomatic incidental finding [11]. Its frequent association with other entities, such as endometriosis, and mimics their symptoms [3,16,17]. The cause of the abdominal bloating and nausea was probably the presence of the pseudo-mass on the pelvis, and since our patient was postmenopausal we did not notice any of the other related symptoms.

Endosalpingiosis has been described in many sites, which include the peritoneum (most common), uterus, urinary bladder, appendix, lymph nodes, skin and abdominal wall [3,5,7,18–21]. This distribution fits with our findings, as almost all of the peritoneal surfaces of the pelvis were covered by small cystic lesions (Fig. 1). It may be very tricky for an experienced surgeon to interpret this condition.

Preoperative diagnosis is unusual. Occasionally, endosalpingiosis presents as multiple pelvic calcifications, and it can have the appearance of metastatic peritoneal implants [22]. Florid cystic endosalpingiosis has been described as

a polypoid multicystic adnexal or uterine mass, and it can therefore be difficult to distinguish it from ovarian or uterine neoplasms in radiological [23] and laparoscopic examinations [7,8]. In our computed tomography scan a pelvic mass appeared with suspected solid areas, probably corresponding to endosalpingiosis calcifications as described above. Due to the suspected diagnosis of recurrent ovarian cancer, we carried out a frozen section examination to assess the situation. It seemed to be endosalpingiosis or maybe borderline ovarian tumor, however, so we proceeded with a complete staging surgery and excision of the lesions, which was justified by the patient's history of ovarian cancer. In addition, we kept in mind that no adjuvant treatment was administered, so the risk of recurrence was probably higher.

Normally, endosalpingiosis does not react with the immunohistochemical stains when using markers such as CA125 or CEA [18,20], and nor do these immunoreactions rise substantially in blood samples [10,24]. This feature could be an additional way to differentiating it from ovarian cancer, although not always in cases of borderline ovarian tumors.

In conclusion, the characteristics of endosalpingiosis make the entity difficult to diagnose and differentiate from an ovarian malignancy. The intraoperative presence of multiple peritoneal cystic lesions, negative tumor markers, and no other evidence of extended malignancy, however, should suggest endosalpingiosis that needs to be confirmed histologically before any additional and more aggressive surgical procedure is undertaken.

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