Case Report

Uterine Serous Cystadenoma or Endosalpingiosis?: A Case Report with a Review of Literature

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Abstract

Endosalpingiosis is a nonneoplastic lesion defined by the presence of tubal epithelium at ectopic sites such as the peritoneum, bladder, appendix, and even uterus. They may be asymptomatic and detected incidentally on ultrasonography. However, cystic endosalpingiosis is also known to be a mimicker of ovarian neoplasms. It is crucial for both the clinician and the pathologist to be aware of this benign lesion so that overdiagnosis and overtreatment can be avoided. We report a case of endosalpingiosis of the uterine serosa in a 45-year-old woman which was misdiagnosed as an adnexal cyst on radiological investigations.

Keywords: *Endosalpingiosis, nonneoplastic, ovarian neoplasm, uterine serosa*

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Introduction

Endosalpingiosis is a benign entity characterized by nonneoplastic proliferation of tubal epithelium. It usually exhibits multiloculated cysts in the peritoneum, urinary bladder, appendix, colon, retroperitoneal lymph nodes, and uterus. It is mostly associated with serous ovarian neoplasms.[1] They are usually asymptomatic and detected incidentally on histopathological examination. Occasionally, they may masquerade as adnexal cysts on radiological investigations.[2]

Very few cases of this entity have been reported to date. We report a case of endosalpingiosis of the uterine serosa in a 45-year-old woman which was misdiagnosed as an adnexal cyst on radiological investigations.

Case Report

A 45-year-old woman presented to the gynecology outpatient department for menstrual complaints. She had irregular cycles with menorrhagia for 5 months. Ultrasound abdomen showed a normal-sized uterus with an intramural fibroid measuring 3 cm in diameter. An adnexal cyst measuring 4 cm in diameter was also seen on the right side. Computed tomography showed an adnexal cyst which was indenting the uterus.

A total abdominal hysterectomy with bilateral salpingo-oophorectomy was done and sent for histopathological examination.

The hysterectomy specimen measured 8 cm × 5 cm × 4 cm. A cyst measuring 5 cm × 4 cm was seen arising from the serosal surface of the uterus. The cyst was uniloculated and filled with serous fluid. The wall thickness of the cyst varied from 0.1 to 0.2 cm. No solid areas or any papillary excrescences were seen. The right-side ovary and tube were seen separate from the cyst thus, ruling out the possibility of the adnexal cyst.

The uterine cavity showed the presence of an intramural leiomyoma of 3.5 cm in diameter with a whorled appearance. The endometrial thickness was 0.3 cm.

Microscopic examination of the cystic structure exhibited a cyst lined by flattened to low cuboidal lining lying against a fibromuscular stroma. The lining was focally ciliated at places [Figure 1]. There were no papillary infoldings, stratification, or any nuclear atypia. The leiomyoma showed whorled architecture and interlacing fascicles of smooth muscle bundles. Based on the above

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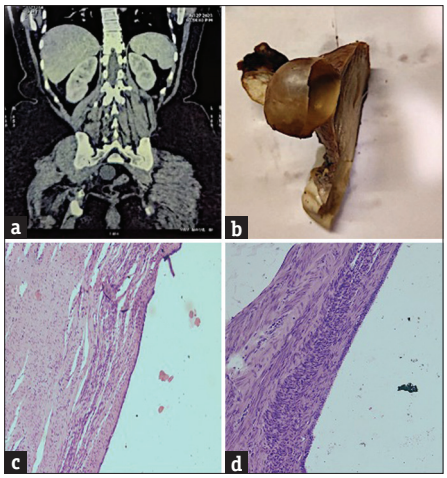

Figure 1: Radiological and pathological fingings of the case. (a) Computed tomography exhibiting a right adnexal cyst which was indenting the uterus. (b) Gross findings exhibiting one half of the hysterectomy specimen with a unilocular cyst measuring 5 cm × 4 cm arising from the serosal surface of the uterus. (c and d) Hematoxylin and eosin-stained sections exhibiting a cyst lined by flattened to low cuboidal ciliated lining lying against a fibromuscular stroma (×40, ×100)

findings, a diagnosis of uterine serosal endosalpingiosis with intramural leiomyoma was made.

Discussion

Endosalpingiosis is a nonneoplastic proliferation of the tubal epithelium at ectopic sites such as the peritoneum, colon, appendix, para-aortic and retroperitoneal lymph nodes and uterus. It usually affects women in the reproductive age group.[2] The possible mechanisms proposed for its pathogenesis include-metaplastic change of the mesothelial lining or the metastatic spread of the epithelium to the ectopic site. The metaplastic change is proposed to be the more common aetiology as pelvic mesothelium is also known to give rise to the Mullerian ducts which eventually form the female genital organs.[3]

Endosalpingiosis was first described by Sampson in 1930 when he found tubal epithelium at ectopic sites in females who had undergone tubectomy. It is usually

Table 1: Clinicopathological features of previously reported cases of uterine endosalpingiosis

Author (year)	Age/sex	Clinical presentation	Radiological findings	Histopathological diagnosis
Singh *et al*.[5]	31/female	Heavy bleeding, lower abdominal pain	USG: Uterus and the right ovary were unremarkable and the left ovary had a multiloculated cyst of 4.3 cm × 3.2 cm	Florid cystic endosalpingiosis
Yığıt *et al*.[6]	44/female	Menorrhagia	USG: A pelvic mass arising from the uterus	Tumor-like cystic endosalpingiosis in the myometrium
Im *et al*.[7]	43/female	Vaginal bleeding	Computed tomography- leiomyoma with cystic degeneration	Intramural florid cystic endosalpingiosis
Shim *et al*.[8]	54/female	Vaginal bleeding	-	Florid cystic endosalpingiosis
Subbaiah *et al*.[9]	53/female	Vaginal bleeding	Contrast-enhanced CT: A 3 cm growth in the endometrium and multiple cysts in the adnexal region and was reported as ovarian cysts	Cystic uterine endosalpingiosis
Heatley and Russell[10]	73/female	Abdominal swelling	-	Florid cystic endosalpingiosis
Scheel *et al*.[11]	48/female	Chronic back pain	USG: Cystic formations with multiple well delimited cysts at the fundus uteri and the adnexa	Cystic endosalpingiosis
Taneja *et al*.[12]	40/female	Chronic back pain and dysfunctional uterine bleeding	CT: Irregular soft-tissue mass in the pelvis and the mass was indenting the posterolateral aspect of the uterus and cervix	Florid cystic Endosalpingiosis
Chang *et al*.[13]	45/female	Intermittent lower abdominal dullness and irregular menstruation	-	Endosalpingiosis of uterine serosa
Hemalatha *et al*.[14]	40/female	Mass per vaginum	-	Cystic endosalpingiosis of uterine parametrium
Yang *et al*.[15]	31/female	An incidental finding of a pelvic mass on sonography	MRI: A single, round trilocular mass with homogeneous cystic content located in the cul-de-sac, which was suspected to be leiomyoma with cystic degeneration	Uterine endosalpingiosis
Carpenter *et al*.[16]	40/female	Pelvic pain, heavy menstrual bleeding	USG: A large, simple-appearing adnexal cyst, presumably originating from the right ovary, along with small fibroids	Uterine serous cystadenoma
Lui *et al*.[17]	47/female	An incidental finding of a right adnexal mass on sonography	USG: A right adnexal mass	Benign mullerian cyst of the uterus

USG: Ultrasonography, CT: Computed tomography, MRI: Magnetic resonance imaging

asymptomatic, however, may present with chronic pelvic pain, menstrual irregularities, or infertility depending on the site. The current case presented with complaints of irregular cycles with menorrhagia. In the present case, it was misdiagnosed to be an adnexal cyst owing to its location. A previous study has reported that endosalpingiosis in premenopausal women predisposes them to a ten times higher risk of malignancy as compared to the normal population.[4]

Although the gross appearance of endosalpingiosis may mimic an ovarian malignancy, the lack of any stratification, nuclear atypia or increased mitotic activity rules out the possibility of a malignant aetiology.[5]

In the majority of cases, endosalpingiosis is seen as multiloculated cysts of variable size. However, in the present case, there was an unilocular cyst filled with thin serous fluid attached to the uterine serosa. Occasionally, it may present as florid cystic endosalpingiosis.

Yığıt *et al*. also reported a case of intramural endosalpingiosis, however, the current case showed origin from the uterine serosa.[6]

Majority of the previously reported cases showed multiloculated cystic masses, however, Lui *et al*. reported a unilocular cyst similar to the present case.[5-17]

The patients presented with complaints of vaginal bleeding, menorrhagia, chronic back pain, and abdominal mass.[5-14,16] In two of the previously reported cases, it was an incidental finding on ultrasonography.[15,17] The present case also presented with complaints of irregular cycles with menorrhagia for 5 months.

Table 1 summarizes the clinicopathological and radiological features of previously reported cases of uterine endosalpingiosis.

Conclusion

It is crucial for both the clinician and the pathologist to be aware of this nonneoplastic entity as it does not warrant a hysterectomy and can be treated conservatively. Awareness about this benign lesion, which is a mimicker of ovarian malignancy, will prevent both misdiagnosis and overtreatment.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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