Florid Cystic Endosalpingiosis of the Uterus
– A Case Report –

Sang Hwa Shim, Han-Seong Kim, Mee Joo, Sun Hee Chang, Ji Eun Kwak

Department of Pathology, Inje University, Ilsan Paik Hospital, Goyang, Korea

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Corresponding Author
Han-Seong Kim, M.D.
Department of Pathology, Inje University, Ilsan Paik Hospital, 2240 Daewha-dong, Ilsan-gu, Goyang
411-706, Korea
Tel: 031-910-7142
Fax: 031-910-7139
E-mail: hs-kim@ilsanpaik.ac.kr

Endosalpingiosis is a non-neoplastic process characterized by benign glandular structures with tubal type epithelium, and usually involves the peritoneum, subperitoneal tissues, and retroperitoneal lymph nodes in women. It is almost always discovered as an incidental finding on microscopic examination without clinical symptoms. Rarely, it forms a large cystic mass which can be misinterpreted as cystic tumors of the uterus or adnexa, which is called “florid cystic endosalpingiosis”. Until now only eight cases have been reported in the literature. The author recently encountered a case of florid cystic endosalpingiosis in the subserosa of the uterine fundus of a 54-year-old female, which was clinically considered to be an ovarian tumor. In this report, the clinical and pathologic features of florid cystic endosalpingiosis of the uterus are described together with differential diagnosis.

CASE REPORT

A 54-year-old woman presented with vaginal bleeding. The ultrasonographic diagnosis was bilateral benign cystic ovarian tumors. The laboratory data showed no abnormalities. Because of a possibility of ovarian cancer, abdominal total hysterectomy and bilateral salpingo-oophorectomy were performed. Intraoperatively, subserosal multicystic polypoid masses in the bilateral uterine horns and posterior fundus of the uterus were found. The bilateral ovaries and tubes were unremarkable. The subsequent course of treatment was uneventful after the operation. Macroscopically, the uterus, measuring 13 × 5 × 4 cm, revealed subserosal cystic polypoid masses in the bilateral uterine horns and the posterior surface of the fundus (Fig. 1). The largest cyst measured 6 × 4.5 × 4.5 cm. On the cut surface, the cysts showed a smooth inner surface and they contained serous fluid. Macroscopically and microscopically, the uterine cervix, endometrium, both ovaries and tubes were unremarkable except for an endocervical polyp. Microscopically, the masses consisted of various sized and shaped cysts. They were lined by a benign-appearing tubal type epithelium surrounded by smooth muscular bundles (Fig. 2). Nearly all of the epithelial lining of the cysts was of the tubular type and was composed of ciliated columnar cells, non-ciliated columnar cells and intercalated peg cells (Fig. 3). The surrounding stroma was predominantly composed of mildly hyperplastic smooth muscle and myofibromatous tissue. Neither endometrial stroma nor periglandular stromal condensa-
tion was found. Some glands were hyperplastic with cellular stratification. No papillary structure or epithelial tufting was found in multiple sections. No mucinous epithelium was observed in the cystic mass. An adenomyotic focus was noted in the fundal myometrium adjacent to the endosalpingiosis. The endometrium was in the normal secretory phase.

**DISCUSSION**

Endosalpingiosis refers to the presence of tubal epithelium outside the uterine tube proper and is generally considered as an incidental finding. However, recently, “florid cystic endosalpingiosis” has been described. Clement and Young described a case of florid cystic endosalpingiosis with tumor-like manifestations, which was characterized by a polypoid mass composed of multiple cysts lined by tubular-type epithelium, hyperplastic smooth muscle and myofibromatous stroma. In addition, a case of florid cystic endosalpingiosis revealed glandular complexity and marked cellular stratification. So-called ‘atypical endosalpingiosis’ was reported in cases of florid cystic endosalpingiosis. In that report, cellular stratification and varying degrees of cellular atypia were found in the endosalpingiosis. In our case, a focus of lesion revealed a stratified tubal epithelium but any cellular atypia was not found.

Of the cystic tumor-like lesions involving the uterus, cystic adenomyosis or endometriosis with tubal metaplasia, adenomatoid tumor and peritoneal inclusion cyst should be differentiated from cystic endosalpingiosis. Cystic endosalpingiosis lacks endometrial stroma in contrast to cystic adenomyosis or cystic endometriosis. Cystic adenomyosis usually contains dark brown cystic fluid. Cystic adenomatoid tumor is macroscopically similar to cystic endosalpingiosis showing uterine cystic masses. Microscopically the cystic spaces are lined with flattened and cuboidal, non tubal type cells admixed with smooth muscle, indicating an adenomatoid tumor of the uterus. Peritoneal inclusion cysts (benign cystic mesotheliomas) are usually adherent to the pelvic organs and they may appear to be a cystic ovarian tumor on the clinical and radiological examinations. Microscopically, however, they are typically lined with a single layer of flat to cuboidal mesothelial cells. In our case, we performed immunohistochemical staining for calretinin to rule out a mesothelial lesion, and the lining epithelium was negative.

The pathogenesis of florid cystic endosalpingiosis is largely unknown. The subserosal location of the cyst in this case may be explained by the so-called ‘secondary Mullerian system theory’. Since conventional endosalpingiosis is usually found in
close proximity to the peritoneum, or the serosal surface or the capsule of lymph nodes, it has been proposed that the coelomic epithelium lining of the peritoneal cavity might undergo a change towards a primary Mullerian epithelium including tubal, endometrial or endocervical epithelium.\textsuperscript{12} However, because the endosalpingiosis can be admixed with small foci of endometrioid glands and endometrial stroma, some have proposed that it could be called “mullerianosis.”\textsuperscript{2,13} Our case revealed a small focus of endometrial glands and stroma adjacent to the cystic endosalpingiosis, supporting the idea that “mullerianosis” could be related to florid cystic endosalpingiosis. Mullerianosis often contains endocervicosis, which was not found in our case.\textsuperscript{14}

In conclusion, endosalpingiosis can rarely undergoes cystic change, resulting in a florid endosalpingiosis associated with clinical manifestations, an intraoperative abnormality, or a striking gross finding. Clinically and radiologically, endosalpingiosis in the uterine horns may simulate a cystic ovarian neoplasm. Thus, awareness of this lesion will facilitate a correct diagnosis by the clinician and pathologist.

\textbf{REFERENCES}