# Florid cystic endosalpingiosis of the uterus

M K Heatley, P Russell

**Abstract**

**A 73 year old woman presented with a right sided adnexal cystic mass. At laparotomy, this proved to be a benign serous ovarian cyst and an aggregation of thin walled subserosal and soft tissue cysts and spongy nodules up to 16 mm in diameter involving the side wall of the uterus and adjacent parametrium. These were removed by total abdominal hysterectomy and bilateral salpingo-oophorectomy. Histologically, the cystic spaces and smaller acini were lined by benign tubo-endometrioid epithelium, with smaller areas typical of serous differentiation and rare microfoci of endocervical-type mucinous epithelium. These features indicated multidirectional Mullerian differentiation in a process that, overall, was consistent with so called florid cystic endosalpingiosis. This lesion is to be distinguished from other benign conditions including multicystic mesothelioma, endometriosis, endocervicosis, florid deep glands of the uterine cervix, and deep Nabothian cysts of the uterine cervix.**

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A 73 year old woman presented with lower abdominal swelling and, at laparotomy, a 70 mm unilocular right ovarian cyst was identified. Contributing to the clinical "mass" were multiple thin walled subserosal cystic spaces and more solid nodules aggregated along the right side of the uterus and in the adjacent parametrial tissue and broad ligament. No other visible lesion was present in or on the pelvic viscera or peritoneum. A total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed. The patient has made an uneventful recovery.

## Pathology

The pathological specimen consisted of an atrophic (73 g) uterus measuring $75 \times 45 \times 30$ mm with attached Fallopian tubes and ovaries, including a unilocular right ovarian cyst and unremarkable left adnexae. The cystic spaces and the nodules, measuring up to 16 mm in diameter, were beneath the uterine serosa (within the superficial myometrium) and extended from the mid corpus to the cervix. The cut surfaces of the nodules revealed a honeycomb pattern of microcysts. Several similar nodules were present in the adjacent right parametrium. An incidental finding was a 12 mm long endometrial polyp.

Histologically, the subserosal, intramyometrial, and parametrial nodules consisted of variably sized and shaped cystic spaces and gland like structures, lined mainly by tubo-endometrioid epithelium (fig 1). Some cysts were lined by serous (tubal) epithelium, whereas others exhibited attenuated serous epithelium or, rarely, endocervical-type mucinous epithelial cells (fig 2). There was no

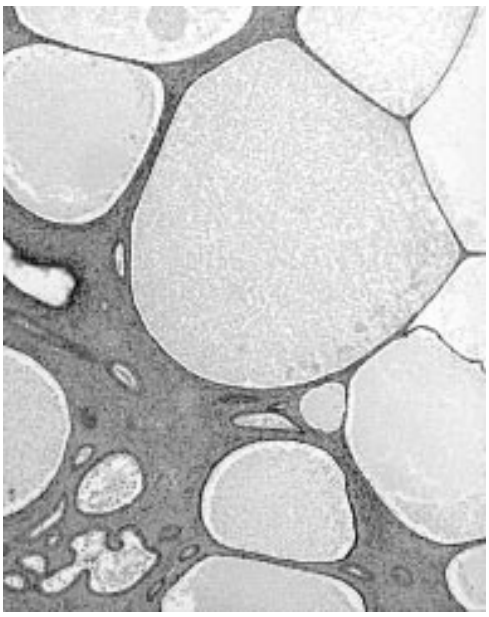

*Figure 1 Florid cystic endosalpingiosis. Variably sized cystic spaces are lined by tubo-endometrial and flattened more serous epithelium. Haematoxylin and eosin stained; original magnification, ×40.*

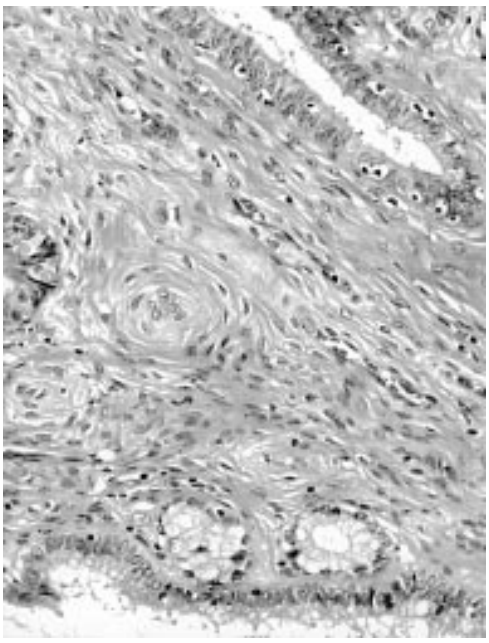

*Figure 2 Florid cystic endosalpingiosis. Variable differentiation is demonstrated with spaces lined partly by mucinous and tubal type epithelium. Original magnification, ×40.*

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evidence of epithelial atypia. The lining epithelium demonstrated intense positivity for cytokeratins (CAM 5.2, MNF 116) and for oestrogen and progesterone receptors. The tubo-endometrioid epithelium also stained strongly for vimentin and CA-125, whereas cytokeratin 5/6 was preferentially demonstrated in the more attenuated serous epithelium. The stroma between the cystic/glandular spaces were fibromuscular without evidence of either endometrial stromal differentiation or recent/old haemorrhage.

There was no evidence of epithelial proliferation in either the endometrial polyp or the right ovarian cyst.

## Discussion

Epithelial structures that occur in women, on or beneath the visceral or parietal peritoneum, in the retroperitoneal lymph nodes, or in the soft tissues of the pelvis and lower abdomen, and with differentiation typical of the lining of the female genital tract, have generically been termed Mullerianosis.[1] They include lesions with tubal/serous differentiation (endosalpingiosis) and the homologous lesions of endometriosis and endocervicosis. Each of these lesions, in its pure form, has a more or less structured anatomical distribution as well as clinical correlates. Mixed forms also occur.[2] The morphological features of our case most closely resemble those recently described by Clement and Young and designated as florid cystic endosalpingiosis,[3] and can be differentiated from the more usual appearance of endosalpingiosis by the fact that tumour like masses were present, with clear intrusion of the process into the subperitoneal connective tissue, as well as the muscular walls of the uterus and other pelvic viscera. In that series of four cases, cysts of up to 42 mm in diameter were described and the lesions were usually lined by tubal type epithelium. Our case may be better termed cystic Mullerianosis because other types of Mullerian epithelium were present, reflecting a multidirectional differentiation.

Although endosalpingiosis can be visualised microscopically in pathology specimens, it is commonly encountered as an incidental finding at laparotomy, on the surface of the pelvic viscera. Tumour like foci of endosalpingiosis have been described rarely in the urinary bladder[2] and in the vermiform appendix.[4]

Of the other non-neoplastic peritoneal lesions of the secondary Mullerian system (Mullerianosis), endocervicosis, which is characterised by a proliferation of mucinous endocervical like glands, can also mimic a neoplasm,[5 6] and has been described in the urinary bladder[5] and the outer wall of the uterine cervix, where the differential diagnosis includes cervical adenocarcinoma. It is of some note that the microfoci of mucinous epithelial differentiation in our case were identified only in the lesions situated in the deep wall of the cervix. This lesion is clearly differentiated from the other major variant of Mullerianosis, namely endometriosis, by the absence of typical cytogenic endometrial like stroma and evidence of a cyclical hormone response (haemorrhage).

Of tumour like lesions arising from the Mullerian duct derivatives, florid deep glands of the uterine cervix can infiltrate the cervical stroma and extend close to the resection surface of the specimen. As in our case, such lesions lack evidence of atypia but, unlike our case, the glands are lined usually by a single layer of tall columnar mucin secreting epithelium and are in continuity with the glands of the endocervical canal,[7] and of course are confined to the deep cervical tissues only. Tubal and tubo-endometrioid metaplasia of the uterine cervix may penetrate deeply into the cervical stroma but should originate from the endocervical canal,[8] rather than arising from or beneath the serosa.

Finally, multiple peritoneal inclusion cysts (benign cystic mesothelioma) are distinguished by the uniform presence of attenuated mesothelial cells lining the cystic spaces, the distribution of the lesions on the surface only of the pelvic structures, and the frequent presence of a mild inflammatory infiltrate in the cyst walls.

The importance of our case, and those few reported in the literature, is that they represent yet another piece in the global "jigsaw" of pathological lesions arising in situ from the secondary Mullerian system.


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