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Case Report

Cystic Endosalpingiosis in Pregnancy –A Case Report

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Abstract: Endosalpingiosis refers to the existence of heterotopic cystic mullarian tissue resembling structures of fallopian tube. It is a rare benign entity whose pathophysiology is not clearly understood. We report a case of cystic endosalpingiosis in a 36-year-old pregnant women during her scheduled lower segment caesarean section. During the procedure the part of uterine fundus and left fallopian tube was studded with multiple cyst like structures, for which left salpingectomy was done. On histopathological examination it was diagnosed as Para tubal cyst with endosalpingiosis. Under recognition of endosalpingiosis may led to its misinterpretation as adenocarcinoma.

Keywords: Endosalpingiosis, mullarianosis in pregnancy, cystic endosalpingiosis.

INTRODUCTION

Endosalpingiosis is the presence of ectopic cystic glands outside the fallopian tube that are lined by fallopian tube type ciliated epithelium [1]. It can be considered as a part of wider group of anomalies of embroyonal origin called mullerianosis [2] consisting in the heterotopic presence of mullarian tissue in pelvic organs or in the distant locations. It occur in pelvic organs including ovaries, fallopian tube serosa, uterine serosa and myometrium or in pelvic peritoneum, it may also occur in bladder or in retro peritoneum and in axillary lymph node.

The most accepted Pathogenesis is the metaplastic change of coelomic epithelium in to tubal like epithelium which is classified as the lesion of secondary mullarian system. This rare gynecological condition usually seen in reproductive age group. It is often seen as incidental finding not usually associated with any pathology. When mass forming, the lesion is called florid cystic endosalpingiosis. This is believed to be caused by distal fallopian tube implants in the ovarian cortex during ovulation. The prevalence of endosalpingiosis is difficult to establish as there are only a few data regarding this.

CASE HISTORY

In A 36- year old female second gravida with no remarkable medical history and an uneventful follow-up attended the hospital unit at term for safe confinement. Since the patient had cephalo pelvic Disproportion, Caesarian section was planned. During lower segment section was noticed to have cysts over uterine fundus, fimbriae and white plaques over fallopian tube for which left partial salpingectomy was done along with LSCS. She had previous history of LSCS and appendicectomy. Patient had no history of infertility treatment or OCP intake. She used to have regular menstrual cycle with normal bleeding. There was no history of dysmenorrhea or dyspareunia. Her blood investigations were within normal limits and radiological evaluation showed live intra uterine gestation and intramural fibroids.

On gross evaluation fallopian tube was dilated which measured 6cm in length and multiple tubal cyst largest measuring 1.2 cm in greatest dimension. On cutting through the cyst extruded serous fluid, no solid area seen. Cut section of fallopian tube was unremarkable. On microscopy observed Para tubal paramesentric cyst (fimbrial cyst), wallthard cell rest and ectopic cystic glands formed by external serous layer, smooth muscle and inner layer of tubal epithelium seen outside the fallopian tube. No atypia or

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proliferative activity is seen. The diagnosis made was fimbrial cyst[1] with endosalpingiosis[2]

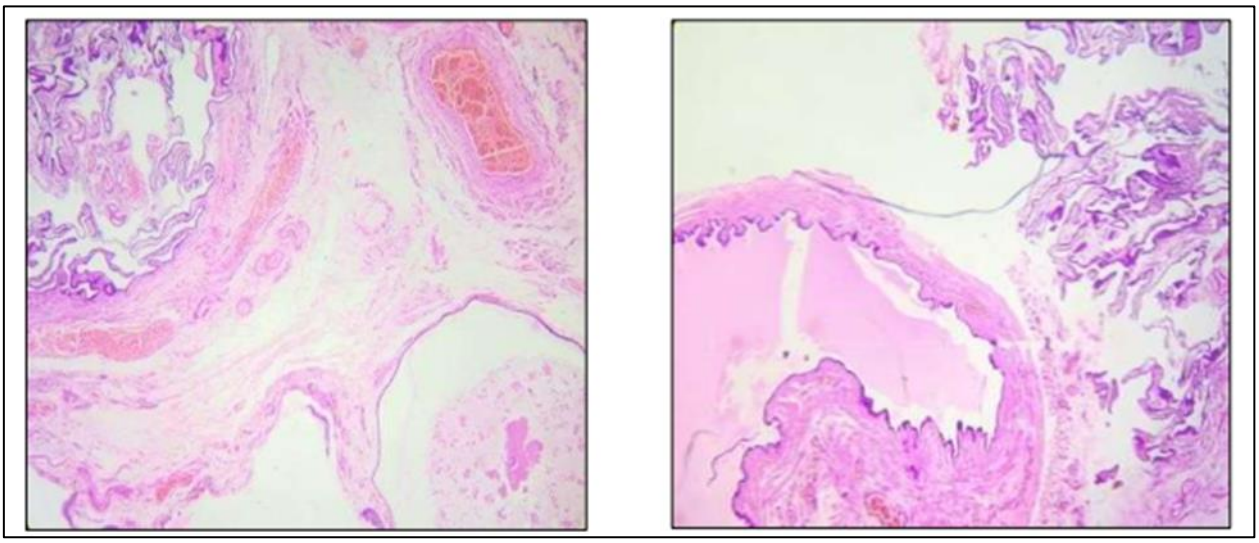

Fig 1 (10x) and fig 2(40x): H & E staining: Ectopic cystic glands outside the FT lined serous layer, smooth muscle layer and inner tubal epithelium

DISCUSSION

Endosalpingiosis was first described by Sampson in 1930; he found epithelium resembling fallopian tube in an ectopic location in women who had undergone previous salpingectomies or tubal sterilization[1]. Endosalpingiosis may be asymptomatic or associated with chronic pelvic pain, dysmenorrhea or menorrhagia. Cystic endosalpingiosis is a part of mullerianosis, disorders consisting in the heterotopic presence of mullarian derived tissues in pelvic organs like uterus, bladder, ovaries, parametrium, uterosacral mesosalpinx, peritoneum and ureters or in distant locations like the small and large intestine (especially in the appendix), coledochal duct, axillary nodes, mediastinum, umbilicus and spine [2]. Although mullerianosis may contain estrogen and progestron receptors, reports of cystic endosalpingiosis and other form of mullerianosis in pregnancy are surprisingly very scarce. They are considered choristomas causing endosalpingiosis, endometriosis, adenomyosis, endocervicosis, and leiomyomatosis peritonealis disseminata.

During organogenesis, a number of genes of the WNT family like WNT4 are activated, producing the necessary signals to conduct the development of the mullerian structures. That is the reason why mutations in the WNT 4 gene cause mullerian duct regression. Recent researches highlighted the possibility that, mullerianosis might be caused by the abnormal reactivation of these genes, causing metaplasia. Another theory states that it would be the presence of remnants of mullerian precursor cells included within the developing tissues. These cells are sensitive to estrogen and progesterone and might proliferate during pregnancy increasing the volume of cyst and thus making them detectable at the end of the pregnancy.

Prentice *et al* reported the rate of previous abdominal and gynecological surgeries as 59.1% and the history of tubal disease was 33.6% [1]. This raises the suspicion that peritoneal implantation may the cause for endosalpingioisis. Our patient had a history of previous appendicectomy and LSCS.Gross appearance of tumor may mimic primary peritoneal tumor or papillary carcinoma of ovary. But absence of mitotic activity or atypia exclude the diagnosis of carcinoma. Endosalpingiosis show positivity with WT1, PAX8, BCL2 & CK7. Calretinin, CK20 and p53 shows negative staining in immunohistochemistry. The development of endosalpingiosis may be hormonally mediated because it is more commonly seen in post-menopausal women.

Martin *et al* at Yale university school of medicine studied tubal epithelial cultures, which demonstrated exhalent proliferation in high estrogen and progesterone state. The possibility of hormonal modulation was further supported by another study in patients with hormonal therapy for endometriosis who also developed endosalpingiosis [3].

Endosalpingiosis is a rare benign entity, resembling peritoneal or ovarian malignancy. The ectopic epithelium in endosalpingiosis will not undergo malignant transformation, it might form large masses mimicking neoplasia. Since endosalpingiosis is sensitive to estrogen and progestron it will enlarge during pregnancy, but did not disappear after pregnancy. Cystic endosalpingiosis is a bengin entity

that should always be considered, even in pregnancy, when it comes to making the differential diagnosis of a pevic or systemic multicystic mass.

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