

## Xanthogranulomatous oophoritis associated with primary infertility and endometriosis

Sir,

Xanthogranulomatous inflammation is a well documented histopathological entity in the gall bladder and kidney. However, xanthogranulomatous inflammation of the female genital tract is unusual and essentially limited to the endometrium. Only a few cases involving the ovary have been reported.<sup>[1,2]</sup> We report a case of a 42-year-old married female who was being investigated for primary infertility for the last 10-12 years. On diagnostic laparoscopy, which was performed 10 years back, endometriosis involving the bladder, uterus and uterosacral ligaments was discovered. Subsequently, the patient developed heaviness and pain in the lower abdomen. She had amenorrhea of four months duration and irregular cycles in the preceding eight to 10 months. Per abdominal examination revealed a midline, non tender, cystic mass arising from pelvis going above umbilicus, about the size of a 26-week uterus. Results of laboratory investigations were – hemoglobin 9.8gm%, total leukocyte count 10,000/cumm, erythrocyte sedimentation rate 58mm in first hour, urine microscopic examination – pus cells 5-6/high power field, urine pregnancy test negative.

Ultrasound revealed a large cystic mass measuring 156x108 mm arising from the pelvis and extending till the epigastric region with homogenous low level interstitial echoes, surrounded by a thick wall, it was suggestive of a right ovarian mass (mucinous cystadenoma). Computerized tomography (CT) pelvis revealed a rounded, aseptate, unilocular cystic mass seen in the right adnexal area with thick enhancing walls and homogenous low density fluid contents reaching up to the level of umbilicus. No mural/luminal solid area/calcification seen. An impression of right ovarian cyst was given. Serum CA-125 level was 81.96U/ml. Polymerase chain reaction for *M. tuberculosis* was negative.

The patient underwent exploratory laparotomy with total abdominal hysterectomy with bilateral salpingo-oophorectomy and omentectomy. Per operatively, a huge ovarian cyst measuring 18x15cm was seen with adhesions to parietal wall and gut loops. Cyst was filled with foul smelling purulent material. Cyst contents were sent for culture and grew *Escherichia coli*.

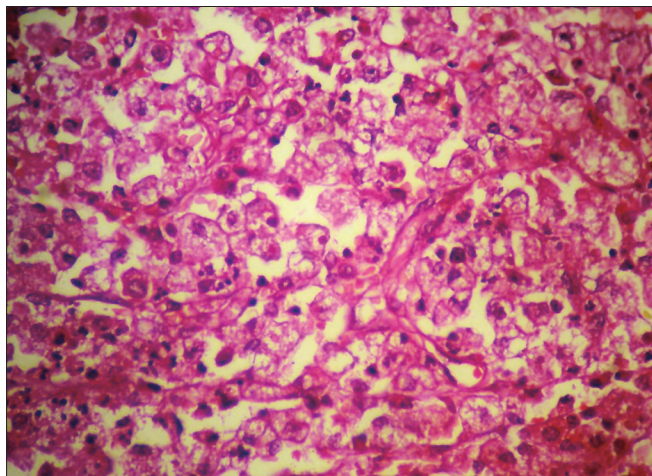
Grossly, the right ovarian cyst measured 14x10x4cm and was

filled with foul smelling, dirty yellowish fluid. Cyst was unilocular and inner lining was grey brown, shaggy with yellowish nodular surface [Figure 1]. Wall thickness varied from 0.5 to 1cm. The specimen of uterus and cervix with left sided adnexa measured 6x5x3cm. On sectioning, two intramural and one subserosal leiomyomata were identified. Microscopic examination of the cyst wall revealed replacement and destruction of ovarian stroma by inflammatory exudate comprising sheets of foamy macrophages, lymphocytes, plasma cells and some neutrophils [Figure 2]. Microsections from bilateral tubes and left ovary were unremarkable. A diagnosis of xanthogranulomatous oophoritis was established.

Xanthogranulomatous inflammation of the female genital tract is very rare and only a few cases of xanthogranulomatous oophoritis have been reported from India.<sup>[3-5]</sup> The involved ovary is usually replaced by a solid, yellow, lobulated mass that is well circumscribed, sometimes involving adjacent organs, thereby mimicking malignancy.<sup>[1,2]</sup>



**Figure 1: Gross appearance of thick walled ovarian cyst showing shaggy, grayish-yellow nodular inner surface.**



**Figure 2: Photomicrograph showing sheets of foamy macrophages interspersed with lymphocytes and few neutrophils. (H and E, 400X)**

Xanthogranulomatous oophoritis has an uncertain etiopathogenesis. Various factors implicated are chronic bacterial infection with *E. coli*, *Proteus* and *Staphylococcus aureus*, inadequate antibiotic therapy, endometriosis and intrauterine contraceptive device.<sup>[3]</sup> In the present case, the culture was positive for *E.coli*. A difficult differential diagnosis is from malakoplakia.<sup>[1]</sup> However, in the index case no Michaelis-Gutmann bodies were found on periodic acid Schiff (PAS) staining. The treatment of choice for xanthogranulomatous oophoritis is oophorectomy.

Since xanthogranulomatous oophoritis is usually associated with pelvic inflammatory disease (PID), endometriosis, intrauterine death etc., these patients should be followed up closely. Although a correct diagnosis is made chiefly through histology, a suggestive preoperative diagnosis of xanthogranulomatous oophoritis could lead to less radical surgery.

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