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Case Report

Serous cystadenoma of ovary with xanthogranulomatous oophoritis: combination of benign ovarian tumour with chronic inflammatory non-neoplastic pseudo-tumour-a rarity

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ABSTRACT

Xanthogranulomatous inflammation is a rare type of chronic inflammation. It destroys the tissue of the affected organ, mostly affecting the kidney and gall bladder. Xanthogranulomatous inflammation of female genital tract being a rare entity, only few numbered cases involving the ovary have been reported in literature. This chronic inflammation of fallopian tubes and ovaries, usually present with pelvic masses, and on imaging and macroscopic appearance, are mostly misdiagnosed as ovarian neoplasm or tuberculosis. We are presenting a case of 14-year-old girl, who presented with an abdominal lump and on clinical and radiological investigations was suspected as ovarian malignancy but histo-pathology reported it as serous cystadenoma with xanthogranulomaotus oophoritis. The rarity of this case lies in the fact that rare destructive inflammatory features was present in a benign neoplasm of the ovary (serous cystadenoma) and that too in a youngest age reported till now.

Keywords: Chronic inflammation, Serous cystadenoma, Xanthogranulomatous oophoritis

INTRODUCTION

Xanthogranulomatous inflammation is a chronic inflammation which is destructive to the affected organ. It is uncommon and non-neoplastic. In this xanthogranulomatous change, the affected organ is infiltrated with foamy histiocytes mixed with multinucleated giant cells, plasma cells, fibroblasts and neutrophils which destroys and replaces the original tissue. Foci of necrosis may also be present in the inflamed tissue.[1,2]

It is commonly found in the kidney, renal pelvis and rarely in organs like ovary and fallopian tube. Other organs in which it has been reported are stomach, ano-rectum, bone, urinary bladder, testes, epididymis, vagina and endometrium. Very few cases of xanthogranulomatous oophoritis have been reported in literature till date. The average age of patients with affected ovaries has been found to be in mid reproductive years (31 years). Usually, this lesion in ovaries and fallopian tube occurs in patients with recurrent pelvic inflammatory disease.

The importance of this case is association of this chronic inflammatory condition with a benign ovarian tumour with no evidence of pelvic inflammatory disease and hence points to a pathogenesis which is very different to what has been speculated till now.

CASE REPORT

A 14-year-old unmarried girl presented in the outpatient department of general gynaecology in AIIMS Patna with

complaints of pain abdomen and abdominal distension for one and a half months. Pain was gradual in onset, mild to moderate in intensity, pricking type and relieved on taking rest. Abdominal distension was gradual in onset over last 1 month. Pain and distension was not associated with any other gastrointestinal or urinary complaints. She attained menarche at the age of 12 years. Her menstruation was normal with mild spasmodic dysmenorrhoea. Her past medical & surgical history was not significant. There was no family history of breast, ovarian or endometrial cancer. On examination, her general survey was normal. Per abdominal examination revealed an abdomino-pelvic lump of 28-week size from right side of pelvis, firm to cystic in consistency, irregular, non-tender, with side to side mobility present (Figure 1).

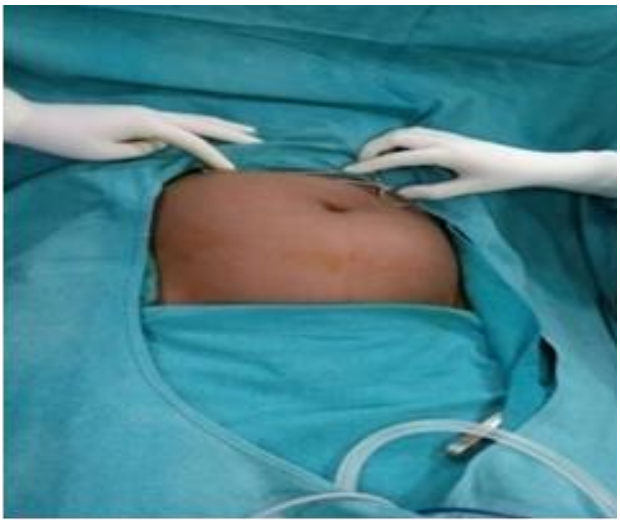

Figure 1: Abdomino-pelvic lump of 28 weeks' size.

On further investigations, trans-abdominal ultrasonography showed a large 19.5 x 11.3, thick walled septated right ovarian cyst (Figure 2).

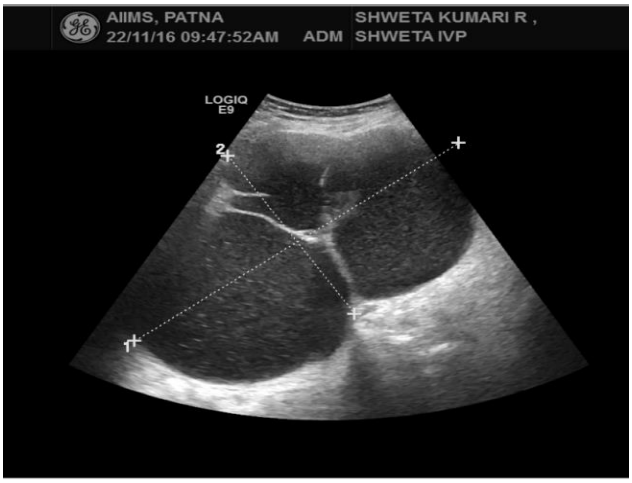

Figure 2: 19.5 x 11.3 cm septated ovarian cyst.

No ascites was reported. Contrast enhanced CT scan of abdomen and pelvis showed a large 19x14x10 cm right adnexal multicystic mass with enlarged lymph nodes around pelvic mass. Right ovary was not seen separately. Moderate right hydroureter and hydronephrosis was seen. Left ovary was 4x3 cm in size and multi cystic. Right para-caval and left para aortic lymph nodes were also seen. Multiple mesenteric lymph nodes were enlarged.

On blood investigations, germ cell tumour markers (AFP, HCG and LDH) were in normal range but serum CA125 was found to be 122 miu/ ml. With a provisional diagnosis of benign ovarian tumour, she was planned for laparotomy. Her pre-operative investigations were within normal limit.

On laparotomy, there was no ascites so peritoneal washing with 50 ml saline was done and fluid was sent for cytology. A right ovarian cyst of size 20 cm x15 cm x10 cm was found with torsion of one and a half turns of pedicle (Figure 3).

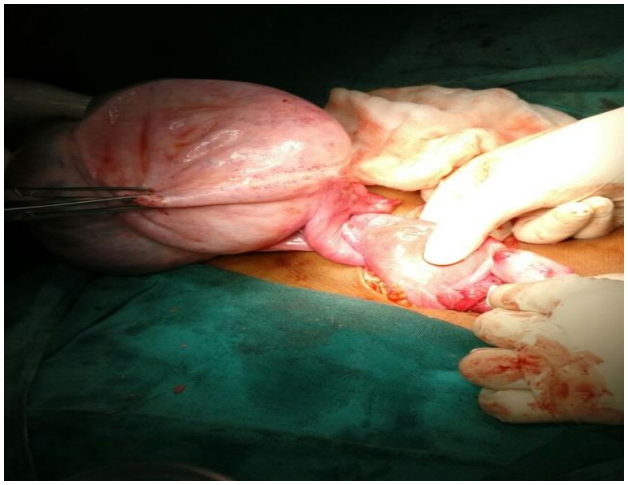

Figure 3: Laparotomy by midline longitudnal incision.

It was multi lobulated and thick walled in places, capsule was intact, and no excrescences were seen on surface. Normal ovarian tissue was not visualised. Right fallopian tube was normal and fimbrial end was stretched over the surface of cyst. Uterus was normal size, central in position. Left ovary was 4x3 cm and multiple cysts of less than a cm was seen over the surface of ovary. Right salpingo- oophorectomy was done. Right external iliac lymph nodes (medial and lateral) were dissected. Para-aortic lymph nodes was found to be palpable 1 cm and left as such. Omental biopsy was also taken.

Exploration of other abdominal organs was done and found to be normal. Sample was sent for histopathological examination and peritoneal cytology. Grossly, cut section of the mass was thick walled, septated, septal thickness was more than 0.5 cm. Foul smelling dark yellowish, tobacco coloured serous fluid came out on puncture (Figure 4). There was a yellowish

solid thickened area identified of size 1.5 x 0.8 cm. Peritoneal fluid cytology showed no malignant cells. Histopathology reported serous cystadenoma with focal xanthogranulomatous oophoritis of right ovary (Figure 5). Lymph nodes showed only reactive lymphadenopathy.

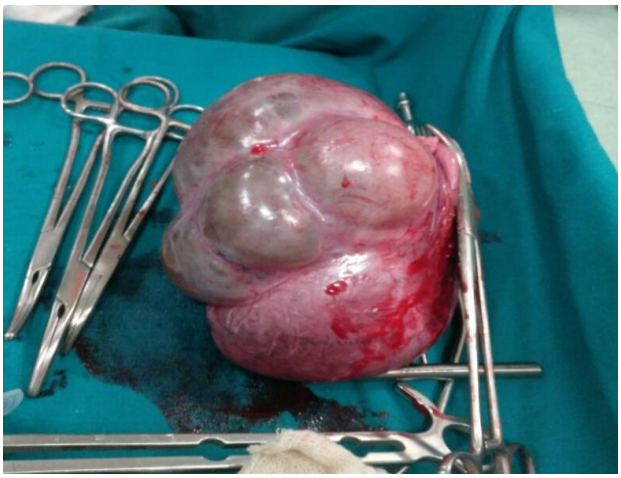

Figure 4: lobulated thick and thin walled ovarian cyst with intact capsule.

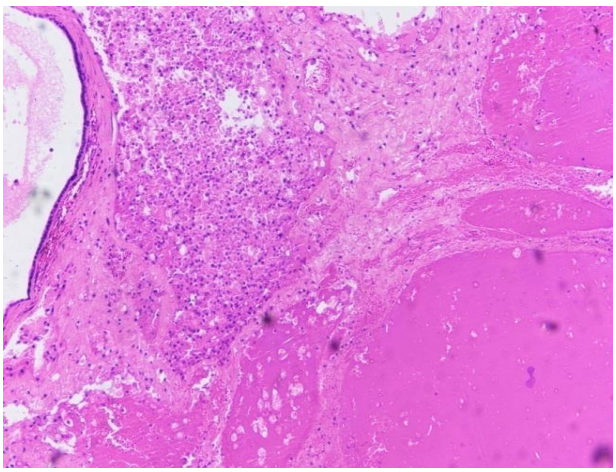

Figure 5: infiltrate of chronic inflammatory cells including foamy histiocytes.

DISCUSSION

Xanthogranulomatous inflammation occurring in female genital tract affects endometrium, fallopian tubes or ovaries. Involvement can be either focal or generalised and forms masses in the pelvic cavity and may invade the surrounding structures as well.[2,3] Kunakemakorn was the first to report inflammatory pseudotumor in the pelvis in serosa of the uterus, left fallopian tube and ovary.[4] Among various literature and case reports, only 16 related cases of Xanthogranulomatous inflammation involving fallopian tube or ovary have been described so far. Only few cases have been reported from India.

Presenting complaints in most are usually lower abdominal or suprapubic pain, fever, menorrhagia, vaginal bleeding, abdominal mass, anemia etc.[5] The clinical pictures, imaging findings and macroscopic observation of xanthogranulomatous oophoritis are confused with ovarian malignancy. But none of the reported cases are in combination with ovarian tumour. In our case, serous cystadenoma was also present.

The involved ovary in each of the previously reported cases was replaced by a solid, yellow and lobulated mass that was well circumscribed sometimes involving adjacent organs, thus simulating malignancy.[6] In our case, lobulated mass was present without involving the adjacent organs. The average age of patients with affected ovaries is 31 years, and the youngest case reported was of 18 years, our case is youngest reported till date being only 14 years of age.[4,7,8] Imaging studies rarely help to reach the correct diagnosis as no specific features of Xanthogranulomatous oophoritis are found except adnexal masses. Presence of non-enhancing intramural nodules in the thickened wall of an ovarian cystic mass may be a unique MR indicator of xanthogranulomatous oophoritis.[1]

This destructive type of chronic inflammation of the affected organs is an uncommon process mostly affecting the kidney. The affected organs show disorganization and infiltration either with focal or sheets of foam cells with other chronic inflammatory cells such as lymphocytes, plasma cells, occasional neutrophils with or without multinucleated or Touton giant cells. The etiopathogenesis of xanthogranulomatous inflammation still remains unclear and a combination of factors seems responsible. Many disorders have the same mechanism of foam cell production. Some have proposed theories like abnormality in lipid metabolism, infection, ineffective antibiotic therapy, and ineffective clearance of bacteria by phagocytes and endometriosis. Sometimes internal organ bleeding and obstruction may predispose to infection, tissue necrosis, followed by the release of cholesterol and other lipids and resultant in phagocytosis by macrophages and formation of foam cells.[9,10]

Gram-negative or anerobic bacteria (*B. fragilis*, *E. coli*, *S. aureus*, *S. typhi* have been considered) commonly implicated in genitourinary tract infections, foreign material such as retained suture material and chronic pelvic inflammatory disease (PID) have been implicated as causative factors.[11] Its association with primary infertility and endometriosis has been reported by Shukla et al.[12]

Xanthogranulomatous oophoritis and salpingitis has also been reported as late sequelae of inadequately treated staphylococcal pelvic inflammatory disease by Punia et al.[13] Differential diagnoses are usually neoplastic, chronic infective and inflammatory conditions. Wather presumed malakoplakia and xanthogranulomatous inflammation as identical chronic inflammatory disease. Lysosomes of phagocytes which fail to digest *E. coli*, get calcified and form concentric calcific bodies (Michaelis-Gutmann

bodies) in malakoplakia. Malakoplakia occurs mainly in the urinary system, but xanthogranulomatous inflammation occurs mainly in the genital system.[14] The treatment for Xanthogranulomatous oophoritis is oophorectomy. Antibiotic therapy has been attempted but it has not succeeded in reducing ovarian mass.[3] Since xanthogranulomatous oophoritis is usually associated with pelvic inflammatory disease, endometriosis, intrauterine device etc., these patients should be followed up closely.

CONCLUSION

Serous cystadenoma of ovary is the most common benign ovarian neoplasm. The etiopathogenesis of serous cystadenoma and xanthogranulomatous oophoritis is not correlated. This case report is special as this rare association is found in a relatively young girl where the usual causative factors of xanthogranulomatous oophoritis like PID and endometriosis was not present. From literature review, it is seen that there is no association of xanthogranulomatous salpingitis or oophoritis with malignancy or radiotherapy. Serous cystadenoma of ovary with xanthogranulomatous oophoritis that was found in our case may be a coincidental finding or may be associated which will be proven in future if we get a number of similar cases.

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