January 2017

Case of xanthogranulomatous oophoritis

Bushra Khan
Aga Khan University, bushra.khan@aku.edu

Aliya Aziz
Aga Khan University, aliya.aziz@aku.edu

Rashida Ahmed
Aga Khan University, rashida.ahmed@aku.edu

CASE REPORT

CASE OF XANTHOGRANULOMATOUS OOPHORITIS

Bushra Khan, Aliya Begum Aziz, Rashida Ahmed*

Department of Obstetrics and Gynaecology, Aga Khan University Hospital, Karachi-Pakistan *Department of Histopathology, Aga Khan University Hospital, Karachi-Pakistan

Xanthogranulomatous inflammation is characterized by destruction of the tissues of the organ involved and replacement by chronic inflammatory cells such as lymphocytes, plasma cells, occasional neutrophils with or without multinucleated or Touton giant cells. Exact aetiology is not known but the theory of infection with organisms like *Proteus*, *E coli*, and *Bacteroides fragilis* is most popular. Xanthogranulomatous inflammation of the female genital tract is not common and usually involves the endometrium; however, xanthogranulomatous inflammation of the ovaries is a rare entity.

Keywords: Lipid laden macrophages; Xanthogranuloma; Chronic inflammation; Oophoritis; Ovaries

J Ayub Med Coll Abbottabad 2017;29(1):162–4

INTRODUCTION

Xanthogranulomatous inflammation is characterized by replacement of normal tissues of the affected organ by lipid containing macrophages with an admixture of lymphocytes, plasma cells and neutrophils. It is an uncommon form of chronic inflammation which mainly involves the kidneys and gallbladder. Other organs in which xanthogranulomatous inflammation has been reported are stomach, anorectal area, bone, urinary bladder and epididymus.[1] Xanthogranulomatous inflammation of the genital tract is not common and usually involves the endometrium, however, xanthogranulomatous inflammation of the ovaries is a rare entity.[2] Kunakemakorn was the first to report inflammatory pseudo-tumour in the pelvis in 1976.[3] Only 29 cases of xanthogranulomatous oophoritis and salpingitis could be tracked through literature survey. A case of xanthogranulomatous inflammation of the ovaries is presented.

CASE

A 47-year-old woman, who had a longstanding history of infertility, was received at the department of Obstetrics & Gynaecology Aga Khan Hospital Karachi. Her only daughter was 22 years of age. She had been having lower abdominal pain and fever for a couple of years.

She was initially diagnosed to have endometriosis for which she was being treated. Her symptoms did not resolve and she had persistent low grade fever. She had loss of appetite and weight. She was evaluated by a physician who prescribed anti-tuberculous drugs and her fever settled. Meanwhile she developed bilateral ovarian masses.

Her investigations showed a haemoglobin level of 12.1 gm/dl, a white blood cell count of 14.8×10^9/l with normal differentials, CA 125 was 12.5 iu/l, and a normal chest x-ray. She had recurrent urinary tract infection and cultures revealed *E coli.*

CT scan pelvis revealed a large complex loculated mass lesion with enhancing septi in bilateral adnexa presenting complex tubo-ovarian masses.

Tubo-ovarian mass lesion in the right adnexa measured approximately 10.8×8.0 cm and in the left adnexa measuring 7.1×3.4 cm. This complex adnexal mass was encasing the left ureter; however, it was not causing hydro nephrosis or hydro ureter. Small bowel loops and rectum were adherent to the mass. No evidence of significant lymphadenopathy was noted. She was prepared for total abdominal hysterectomy, bilateral removal of tubo-ovarian masses and possible gut resection. Prior to surgery bowel preparation was done. Keeping in view the CT scan report stating that mass was encasing the ureters, prior to surgery bilateral ureteric stents were placed which were removed later.

Midline incision was given. There were massive adhesions of bowel with the tubo-ovarian masses. Biopsy from left tubo-ovarian mass was sent for frozen section and it revealed chronic inflammatory tissue. Total abdominal hysterectomy along with removal of tubo-ovarian masses was only possible after general surgeon resected the involved small bowel and made ileostomy. Specimen was sent for histopathology. Patient had an uneventful recovery and hospital stay. She was discharged on 7th postoperative day.

Histopathological findings are shown in panel-1.

Gross pathology: The uterus measured 8.4×4.6×3 cm with smooth endometrium. The left ovarian mass measured 7×5×3 cm with rugged outer surface. Ovarian tissue was torn at various places probably representing adhesions with the surroundings. Right tubo-ovarian mass was 10×7×5 cm with breached capsule. On serial sectioning, there were grayish-white and necrotic mass with focal hemorrhage and variable-thickness pseudo-capsule.

Microscopic examination: All specimens were fixed in 10% formalin, dehydrated in descending grades of alcohol, sections were paraffin embedded and processed for histologic and immunohistochemical stains. Sections from endometrium revealed tubular to tortuous endometrial glands lined by tall columnar cells exhibiting nuclear pseudo stratification. Stroma was spindle shaped and cellular. Left ovarian mass showed multiple corpora albic antes and mild stromal hyperplasia. Left fallopian tube showed hydro salpinx and endometriosis. The pathological changes in the right tubo-ovarian mass exhibited dense acute and chronic inflammation and abundance collection of lipid laden macrophages along with necrosis. The inflammatory cells were mainly lymphocytes admixed with plasma cells, small number of neutrophils and eosinophils. The foamy histiocytes were identified with abundant cytoplasm, small lipid vacuoles and hypochromatic nuclei (Figures-1 and 2). The fibrous pseudocapsule was infiltrated with chronic inflammatory cells.

Immunohistochemistry showed positive staining for CD68 while CKAE1/AE3 was negative. Periodic acid-schiff (PAS) and acid fast stains were negative. The pathological diagnosis was xanthogranulomatous oophoritis.

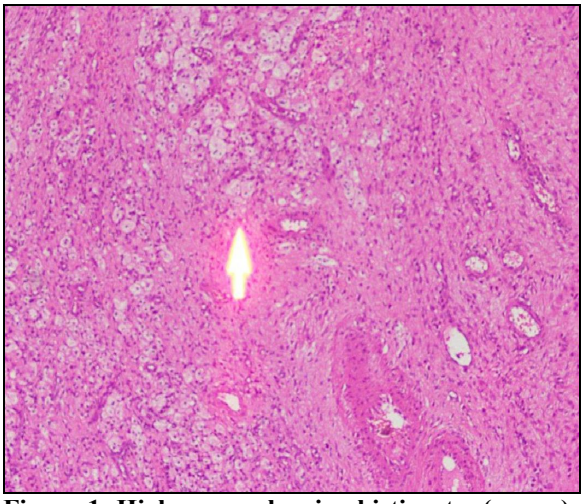

Figure-1: High power showing histiocytes (arrow)- 10×10 H and E Stain.

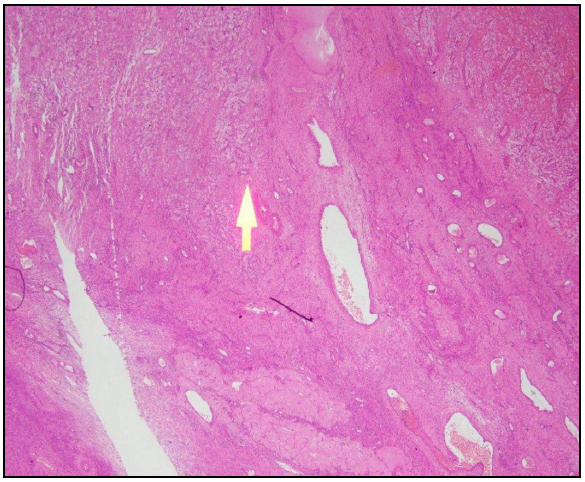

Figure-2: Histiocytes (arrow) in Xanthogranulomatous oophoritis. Corpora albicantes at the bottom. 4×10 H and E Stain.

DISCUSSION

Xanthogranulomatous inflammation is characterized by destruction of the tissues of the organ involved and they are replaced by chronic inflammatory cells such as lymphocytes, plasma cells, occasional neutrophils with or without multinucleated or Touton giant cells. Xanthogranulomatous inflammation of the female genital tract mostly affects the endometrium however we think that there is ample evidence that it equally involves the ovaries.[1,3]

This is a disease of reproductive age. The average age is 35.2 years (range 2–47). The youngest documented case report of xanthogranulomatous inflammation of the ovaries is of a 2-year-old girl.[4] Another girl who was just 18 years of age was also reported to have xanthogranulomatous inflammation of the ovaries.[5]

The pathogenesis of xanthogranulomatous oophoritis is unclear and many theories that are of aetiopathogenesis have been postulated. They are theories of: infection, endometriosis, intrauterine contraceptive device, inborn errors of lipid metabolism, and drug induced. Amongst these theories, the most accepted theory is of infection, which is supported by clinical evidence of infection and growth of bacteria such as *Escherichia coli, Bacteroides fragilis*, and *Proteus vulgaris* from the affected tissue by culture.[6]

Patients usually present with a long-standing history of pelvic inflammatory disease and suffer symptoms like anorexia, fever and lower abdominal pain. There is a long history of antibiotic intake as well. It has been observed that usually patients with xanthogranulomatous oophoritis have low parity or infertility.[7] Clinical manifestations and imaging findings often lead to the diagnosis of either a tubo-ovarian mass or a malignancy.[8] However cases of

xanthogranulomatous oophoritis associated with premature ovarian failure, bowel obstruction, diverticulitis and occurrence after typhoid and uterine artery embolization have been reported.[9]

Tubo-ovarian abscesses are frequently misdiagnosed as ovarian malignancies due to their unusual appearances at computed tomography (CT) and magnetic resonance (MR) imaging.[10] In our case the differential diagnosis was endometriosis and ovarian malignancy. Our patient had history of infertility and was being treated for endometriosis and tuberculosis in the past; however, CT scan had favoured the diagnosis of tubo-ovarian abscess. In most of the cases mass is unilateral, 3–7cm in maximum diameter with capsule adherent to surrounding structures like fallopian tubes, uterus and bowel. The cross section is solid with areas of haemorrhage, necrosis and purulent collection. Frozen section was carried out in cases which mimicked ovarian malignancy. Histopathology shows sheets of foamy histiocytes, ill formed granulomas, multinucleated giant cells and inflammatory cells. This fibrous organization of the chronic inflammatory process is probably responsible for the pseudo-tumoural appearance on gross examination. Because of the presence of foamy histiocytes, malakoplakia may also be considered in the differential diagnosis. Malakoplakia and xanthogranulomatous inflammation probably share a common pathogenetic mechanism.[11] Malakoplakia is thought to represent an inflammatory process in which macrophages do not have the ability to kill the bacteria after phagocytosis. Microscopically the lesion is composed of histiocytes with eosinophilic granular cytoplasm (von Hansemann histiocytes). Malakoplakia characteristically shows the cytoplasmic concentric calcific bodies known as Michaelis-Gutmann bodies which are absent in xanthogranulomatous inflammation. Immuno-histochemical stains are helpful in establishing the diagnosis, including CD68 for histiocytes, CD 20 for B lymphocytes, CD 3 for T lymphocytes.

CONCLUSION

Xanthogranulomatous oophoritis is a rare entity which poses a diagnostic challenge for gynaecologists, radiologists and histopathologists. It must be considered in the differential diagnosis of ovarian mass in patients with a history of pelvic inflammatory disease. Radiological findings mimic ovarian malignancy. Frozen section can rule out malignancy and avoid extensive surgery. Careful histopathology by experienced pathologists aided by immunohistochemistry can lead to a definitive diagnosis.

REFERENCES

1. Bindu SM, Mahajan MS. Xanthogranulomatous oophoritis: A case report with review of literature. Int J Health Allied Sci 2014;3(3)187–9.
2. Chechia A, Bahri N, Felah R, Khaireddine A, Sakouhi M, Zakhama A. Tubo-ovarian xanthogranulomatous inflammation. Report of a case. Tunis Med 1999;77(11):593–6.
3. Kunakemakorn P, Ontai H, Balin H. Pelvic inflammatory pseudotumor: A case report. Am J Obstet Gynecol 1976;126(2):286–7.
4. Tanwar H, Joshi A, Wagaskar V, Kini S, Bachhav M. Xanthogranulomatous salpingooophoritis: The youngest documented case report. Case Rep Obstet Gynecol 2015;2015:237250.
5. Shilpa D, Sulhyan K, Sachin B, Gosavi A, Ramteerthkar N. Xanthogranulomatous oophoritis: Case report. Indian J Basic Appl Med Res 2013;2(7):745–9.
6. Gray Y, Libbey NP. Xanthogranulomatous salpingitis and oophoritis:A case report and review of the literature. Arch Pathol Lab Med 2001;125(2):260–3.
7. Kishor HS, Rajshri PD, Dravid NV. Xanthogranulomatous oophoritis mimicking as an ovarian neoplasm. J Case Rep 2014;4(1):100–3.
8. Sharma S, Phadnis P, Kudva R, Valiathan M, Shetty J. Xanthogranulomatous salpingo-Oophoritis presenting as a tubo-Ovarian mass-A case report with brief review of literature. Int J Health Sci Res 2016;6(3):316–9.
9. Son J, Raetskaya-Solntseva O, Tirman PA, Water GS, Kelly MG. Xanthogranulomatous oophoritis presenting as an adenexal mass and bowel obstruction. Acase report. J Reprod Med 2015;60:273–6.
10. Kim SH, Kim SH, Yang DM, Kim KA. Unusual causes of tuboovarian abscess: CT and MRI Imaging Findings. Radiographics 2004;24(6):1575–89.
11. Walther M, Glenn JF, Vellios F. Xanthogranulomatous cystitis. J Urol 1985;134(4):745–6.

Received: 16 September, 2016	*Revised: 11 November, 2016*	*Accepted: 6 December, 2016*

Address for Correspondence:
Dr. Bushra Khan, Department of Obstetrics and Gynaecology, Ayub Teaching Hospital Abbottabad-Pakistan
Cell: +92 332 891 7837
Email: alrazimedicalstore@yahoo.com, Bushra.khan@aku.edu