



### Case Report

## A Rare Xanthogranulomatous Oophoritis Presenting as Ovarian Cancer

### Authors

**Mahesh Kalloli, Uttam D Bafna, Geetashree Mukherjee, Uma Devi K, Gurubasavangouda, Praveen S Rathod,**  
Department of Gynaecologic Oncology, Kidwai Memorial Institute of Oncology, Bangalore

### Address for Correspondence

**Dr. Mahesh Kalloli,**

No. 5, AB Type, Block-1,

Kidwai Memorial Institute of Oncology campus,

Dr MH Marigowd Road,

Bangalore-29.

**E-mail:** mahesh.kalloli@gmail.com

### Citation

Kalloli M, Bafna UD, Mukherjee G, Devi UK, Gurubasavangouda, Rathod PS. A Rare Xanthogranulomatous Oophoritis Presenting as Ovarian Cancer. *Online J Health Allied Scs.* 2012;11(2):11. Available at URL:<http://www.ojhas.org/issue42/2012-2-11.htm>

### Open Access Archives

<http://cogprints.org/view/subjects/OJHAS.html>

<http://openmed.nic.in/view/subjects/ojhas.html>

Submitted: Apr 20, 2012; Accepted: Jul 3, 2012; Published: Jul 25, 2012

**Abstract:** Xanthogranulomatous inflammation is an uncommon form of chronic inflammation that is destructive to affected organs; it is characterized by the presence of lipid-filled macrophages with admixed lymphocytes, plasma cells, and neutrophils. Only a few cases of xanthogranulomatous oophoritis have been reported to date. We describe a rare case of xanthogranulomatous oophoritis with involvement of omentum.

**Key Words:** Xanthogranulomatous Oophoritis; Ovarian Cancer; Omentum

### Introduction

Xanthogranulomatous inflammation is a special form of chronic inflammation that is destructive to normal tissue of affected organs.<sup>1</sup> This is an uncommon process mostly affecting the kidney.<sup>2</sup> However, other organs in which xanthogranulomatous inflammation has been reported are the gallbladder, stomach, anorectal area, bone, urinary bladder, testis, epididymis, vagina and endometrium. Xanthogranulomatous inflammation of the female genital tract is unusual and is essentially limited to the endometrium.<sup>3</sup> Only a few cases involving the ovary have been reported.<sup>3,4</sup> It is characterized by a massive infiltration of the tissues by lipid-laden histiocytes admixed with lymphocytes, plasma cells and polymorphonuclear leucocytes. There is no minimal amount requirement for the histiocytes to make this diagnosis. The pathogenesis is unclear. The proposed causes are infection, ineffective antibiotic therapy, abnormality in lipid metabolism, endometriosis, or ineffective clearance of bacteria by phagocytes. A combination of these factors may be responsible. This lesion occurs in patients with recurrent pelvic inflammatory disease.<sup>4</sup>

Clinically and radiologically, Xanthogranulomatous inflammation is mimicking tumor of the ovary and fallopian tube. Kunakemakorn was the first to report inflammatory pseudotumor in the pelvis in 1976.<sup>5</sup> To date, only 13 cases of xanthogranulomatous inflammation in the ovary and fallopian

tubes.<sup>6</sup> In this paper we report a rare case of xanthogranulomatous oophoritis with involvement of omentum with review of the literature on this subject.

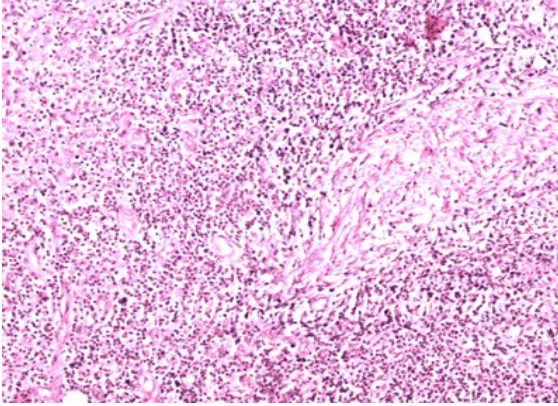
### Case Report

A 45 year old woman, married and a mother of two children, presented with 3 months history of intermittent severe sharp abdominal pain localized to the left suprapubic region and low backache. She had had an intrauterine device (IUD) for 4 years and experienced an episode of pelvic inflammatory disease after the IUD was first inserted. The patient's medical history was otherwise unremarkable. Her physical examination revealed a large mass in left iliac fossa with restricted mobility.

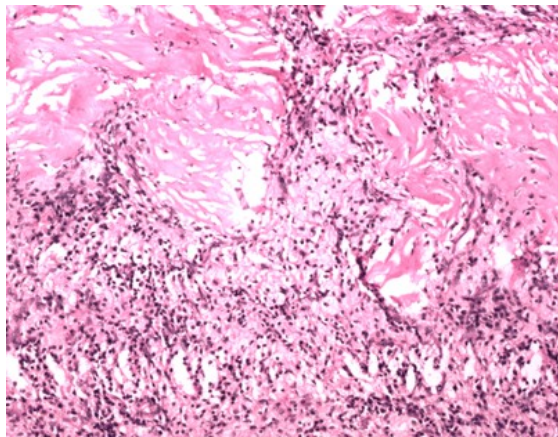
Laboratory investigation revealed a normal hemogram and normal liver and renal function and normal levels of CEA and CA-125. Pelvic ultrasound showed a large lobulated lesion in left adnexa. MRI abdomen and pelvis shown a large 8.7×5.1×7.7 cm enhancing mass lesion involving the left ovary with peri ovarian enhancing fascial thickening involving the adjacent uterus, bladder and peri rectal fascia with left sided hydronephrosis which was thought to represent either an inflammatory mass or an ovarian neoplasm.

Patient was taken for exploratory laparotomy. During the surgery, it was noted that the left adnexal structures were involved with an 8 to 8 cm mass adherent to omentum. Frozen section report came as inflammatory lesion with fibrosis. The patient underwent a total abdominal hysterectomy and bilateral salpingo-oophorectomy with total omentectomy. The patient had an uneventful recovery and was discharged on the postoperative day 6.

The histopathology of the surgical specimens revealed Xanthogranulomatous inflammation of ovary and omentum (Figure 1 and 2). Microscopy showed Patient was referred to urologist for left ureteric stenting. Now patient is on regular follow up.



**Figure 1: Microscopic picture showing ovarian structure in the background of dense chronic inflammatory cells (hematoxylin-eosin)**



**Figure 2: Lipid-containing macrophages and mixed inflammatory infiltrate (hematoxylin-eosin).**

### Discussion

Xanthogranulomatous inflammation is a special form of chronic inflammation that is destructive to normal tissue of affected organs.<sup>1</sup> This is an uncommon process mostly affecting the kidney.<sup>2</sup> The affected organs suffer disorganization and infiltration with focal or sheets of foam cells admixed with chronic inflammatory cells such as lymphocytes, plasma cells, occasional neutrophils with or without multinucleated or Touton giant cells. Xanthogranulomatous inflammation occurring in female genital tract affects endometrium, fallopian tubes or ovaries focally or entirely, which clinically forms mass like lesion in the pelvic cavity and invades the surrounding tissues. Only 13 related cases of Xanthogranulomatous inflammation involving fallopian tube or ovary have been described so far. Only a few cases have been reported from India.<sup>7</sup> The clinical manifestation, imaging detection and macroscopic observation of xanthogranulomatous oophoritis are subject to be confused with ovarian malignancy. Eight reported cases as well as ours were all misdiagnosed as ovarian cancers.

The average age of patients with affected ovaries is 31 years. The involved ovary in each of the previously reported cases was replaced by a solid, yellow, lobulated mass that was well circumscribed and consisted of xanthogranulomatous inflammation.<sup>8</sup> Presenting complaints were lower abdominal or suprapubic pain (sometimes unilateral), fever, menorrhagia, or vaginal bleeding. On physical examination, there was adnexal tenderness and a pelvic mass. The pathogenesis of xanthogranulomatous inflammation remains unclear. Many unrelated disorders may have the same mechanism of foam cell production<sup>9</sup>, proposed causes are infection, ineffective antibiotic

therapy, abnormality in lipid metabolism, endometriosis, and ineffective clearance of bacteria by phagocytes. A combination of factors may be responsible. For example, bleeding and obstruction may predispose to infection, tissue necrosis occurs, followed by the release of cholesterol and other lipids and phagocytosis by macrophages.<sup>9</sup>

Wather<sup>10</sup> presumed malakoplakia and xanthogranulomatous inflammation were identical chronic inflammatory disease. In malakoplakia, the cytoplasmic concentric calcific bodies (Michaelis-Gutmann bodies) are phagolysosome of phagocytes which calcifies after phagocytes phagocytose *E. coli* but fail to digest it. Malakoplakia occurs mainly in the urinary system, but xanthogranulomatous inflammation occurs mainly in the genital system.

Presence of nonenhancing intramural nodules in the thickened wall of an ovarian cystic mass may be a unique MR indicator of xanthogranulomatous oophoritis. Although a correct diagnosis is made chiefly through histology. Xanthogranulomatous oophoritis is often misdiagnosed by pathologists if they don't keep this entity in mind. This may be due to the rarity of the condition. If the lesion is mainly focal scattered lymphocytes, it may be misdiagnosed as secondary lymphoma or leukemia. If the lymphocytes are scattering diffusely and foam cells are seldom, a diagnosis of malignant small cell tumor with stromal luteinization may be rendered. If there are small amount of obvious fibrosis and foam cells, a diagnosis of sclerosing stromal tumor may be made. However, the right diagnosis is possible to be made as long as pathologists elevate vigilance and master the pathological features. Immunohistochemical stains are helpful in establishing the diagnosis, including CD68 (foam cells positive), CD3 (T lymphocyte marker), CD20 (B lymphocytes marker), ? and ? (both positive in polyclonal B lymphocytes).

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.

### References

1. Franco V, Florena AM, Guarneri G, Gargano G. Xanthogranulomatous salpingitis: case report and review of the literature. *Acta Eur Fert.* 1990;21:197-199.
2. Goodman M, Curry T, Russell T. Xanthogranulomatous pyelonephritis (XGP): a local disease with systemic manifestations: report of 23 patients and review of literature. *Medicine.* 1979;58:171-181.
3. Jung SE, Lee JM, Lee KY, Han KT, Hahn ST. Xanthogranulomatous oophoritis: MR imaging findings with pathologic correlation. *Am J Roentgenol* 2002;178:749-751.
4. Gray Y, Libbey NP. Xanthogranulomatous salpingitis and oophoritis: a case report and review of the literature. *Arch Pathol Lab Med* 2001;125:260-263.
5. Kunakemakorn P, Ontai H. Pelvic inflammatory pseudotumor: A case report. *Am J Obstet Gynecol.* 1976;26:286-287.
6. Zhang XS, Dong HY, Zhang LI, Desouki MM, Zhao C. Xanthogranulomatous Inflammation of the Female Genital Tract: Report of Three Cases. *J Cancer* 2012;3:100-106.
7. Shukla S, Pujani M, Singh SK, Pujani M. Xanthogranulomatous oophoritis associated with primary infertility and endometriosis. *Indian J Pathol Microbiol* 2010;53:197-198.
8. Clement PB. Nonneoplastic lesions of the ovary. In: Kurman RJ, ed. *Blaustein's pathology of the female*

- genital tract*, 4th ed. New York: Springer-Verlag, 1994. pp599–604.
9. Cozzuto C, Carbone A. The xanthogranulomatous process: xanthogranulomatous inflammation. *Pathol Res Pract*. 1988;183:395–402.
  10. Walther M, Glenn JF, Vellios F. Xanthogranulomatous cystitis. *J Urol*. 1985;134:745-746.