

BILATERAL XANTHOGRANULOMATOUS SALPINGO-OOPHORITIS

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OBJECTIVE

Xanthogranulomatous salpingo-oophoritis (XGSO) is a rare histological diagnosis resulting from a chronic inflammatory process leading to destruction of normal healthy tissue; histologically confirmed by the presence of lipid-rich foamy macrophages. Clinically and radiologically, xanthogranulomatous benign tumours mimick other pelvic pathologies – namely pelvic inflammatory disease (PID) and malignancy. The literature describes cases of XG diagnoses predominantly within the kidneys and gallbladder. In this report, we present a patient with bilateral xanthogranulomatous salpingo-oophoritis.

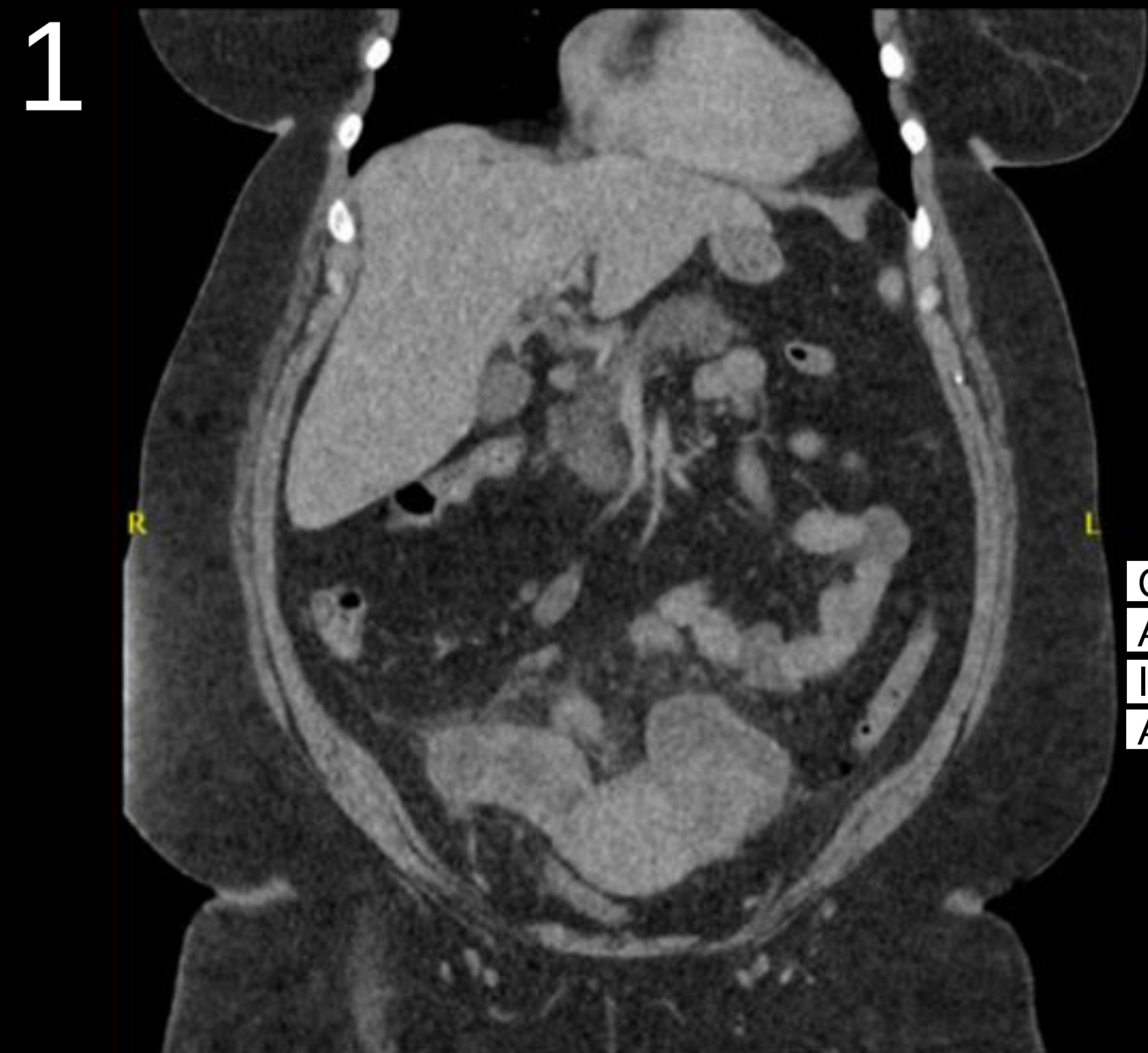
CASE REPORT

This care report describes a 51-year-old lady, G3 P1+2, who presented to the surgical assessment unit with severe abdominal pain, acute urinary retention, constipation, and fever for several days, with a raised CA-125. This patient had an extensive gynaecological history of menorrhagia – managed unsuccessfully with endometrial ablation, previous c-section, recurrent PID and consistently problematic intrauterine contraceptive use (with multiple A+E attendances).

CASE REPORT
CONTINUED

A surgical review suspected intra-abdominal pathology, and a CT scan (figure 1) of the abdomen and pelvis was arranged which showed bilateral adnexal masses with possible tubo-ovarian abscesses – suspicious of severe PID or a malignant aetiology. Blood tests revealed a raised white cell count (WCC) 16.0, neutrophils 12.60, haemoglobin (Hb) 131, platelets 334, INR 123, CRP 256, normal renal function, and a cancer antigen 125 (CA-125) of 132.2 (high).

Discussion within MDT decided to manage her conservatively as per the PID protocol with antibiotic therapy, removal the IUS and a follow-up MRI scan. This MRI scan (figure 2) showed persistent bilateral adnexal masses with fistulation into the sigmoid colon (figure 3) which prompted review by the colorectal surgical team. This resulted in a joint operation between gynaecology and colorectal surgeons for a total abdominal hysterectomy, bilateral salpingo-oophorectomy, bowel adhesiolysis and resection. Histopathologically, the appearances visualised both macroscopically and microscopically are consistent with a diagnosis of uterus and cervix adenomyosis plus bilateral xanthogranulomatous salpingo-oophoritis, with no evidence of malignancy. The main challenges faced in this case was the uncertainty of the diagnosis, the possible requirement for an extensively invasive operation in a young woman and the large BMI of the patient restricting the use of ultrasound scanning.



DISCUSSION

XG inflammation is a recognised and well-documented histopathological diagnosis within the gallbladder and kidney. Following a thorough literature review, there were very few cases reported within the female genital tract, and even less so as a bilateral presentation. Kunakemakorn et al published the first report of pelvic XG pseudotumours and Chiesa-Vottero published a report that between the years of 2003 and 2018, there were 13 recorded cases of xanthogranulomatous salpingitis.

The clinical and radiological signs of XG may vary, leading to easy misdiagnoses and incorrect treatment plans formulated. With differentials of PID or malignancy, the confirmatory diagnosis is either delayed or 'overly-invasive', respectively. The risk factors for XG include endometriosis, leiomyoma, inadequately treated PID, IUD in situ, abnormal lipid metabolism, immunosuppression, previous abdominal surgery, diabetes mellitus type 2, hyperlipidaemia, and untreated urinary tract infections (particularly pathogens like Escherichia coli, Proteus spp, Staphylococcus aureus, Bacteroides fragilis, and Salmonella typhi).

From review of the published reports and our case, some clinical and pathological features can be surmised: (1) female genital tract XG inflammation is primarily unilateral, affecting either the ovaries or fallopian tubes; (2) presentation following chronic history of PID; (3) between the ages of 21 to 75, with a mean age of 45 years, (4) initial symptoms of abdominal pain, anaemia, anorexia, fever, irregular colporrhagia, urinary retention, or infertility with examination findings of adnexal mass(es) with associated tenderness, (5) laboratory findings reveal an elevated ESR and WCC count, (6) imaging studies (ultrasound scan, CT or MRI) display a well-defined solid mass, commonly misdiagnosed as a malignant tumour or metastases.

CONCLUSION

This case supports other literature, suggesting that a high index of investigative suspicion is required due to the non-specific presenting features. For future research and clinical purposes, a clearer understanding of the clinical complaints, MRI findings, and patient risk factors may improve diagnostic abilities and guide appropriate management.

Diagnostically, our recommendation is that performing stains for gram stain, acid-fast bacilli and fungi should be executed post-operatively. Intra-operatively, utilising the frozen section technique and sending cultures, bloods, urine cultures will help aid diagnosis, potentially reduce the requirement of extensive surgery, and help to prescribe targeted antibiotic therapy if pathogens are isolated. With the mean age of diagnosis being 45-years, and the current management is radical in comparison to the oophorectomy recommended for this condition. Considering the PID risk factors, potentially long-term follow up for these patients with their GPs or gynaecology department should be considered to assess for the development of XG changes. This case further highlights the emphasis for early consultation with MDT members to deliberate surgical options. This patient endured extensive surgery, so in the future it may be pertinent to consider a diagnosis of chronic inflammatory changes based on the bilateral nature of disease, risk factors and MRI findings.

