

Rare case of non-healing episiotomy wound: a diagnostic challenge

R Nagpal^a, K Kshitija^b, V V Sreedhar^c

Abstract

We report a case of 28-year old post partum woman who presented in our hospital with the complaints of a non healing episiotomy wound and fever. Examination revealed gaping of the episiotomy wound and a small mass with no active vaginal bleeding. Laboratory investigations showed low haemoglobin, mildly raised total leucocyte count and raised ESR. Histologically it was well characterized by infiltration of the tissue by lipid-laden histiocytes and inflammatory cells and was suggestive of xanthogranulomatous inflammation. It usually poses diagnostic and therapeutic challenges for the clinician. To the best of our knowledge xanthogranulomatous inflammation associated with the non-healing episiotomy wound has never been reported.

Key words: xanthogranulomatous inflammation, lipid laden histiocytes, episiotomy

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Introduction

Xanthogranulomatous inflammation (XGI) is a rare chronic inflammation characterized by the presence of a large number of lipid-containing macrophages with an admixture of lymphocytes, plasma cells, and neutrophils. Multinucleated giant cells may also be present. This uncommon process is best known to

occur in the kidney. Other organs in which XGI has been reported are the gallbladder, stomach, anorectal area, bone, urinary bladder, testis, epididymis¹. Only few cases of XGI of female genital tract (FGT) have been reported till date. According to our research XGI of the vagina with the non-healing episiotomy wound has not yet reported.


^a Assistant Professor, Department of Pathology, Bhaskar Medical College and General Hospital, Hyderabad-500075, Telangana, India.

^b Assistant Professor, Department of Pathology, Bhaskar Medical College and General Hospital, Hyderabad-500075, Telangana, India.

^c Professor, Department of Pathology, Bhaskar Medical College and General Hospital, Hyderabad-500075, Telangana, India.

Correspondence: KK, e-mail: <Kshiti80@yahoo.co.in>

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 <https://orcid.org/0000-0003-4153-0748>

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Case report

A 28-year old, with one living child, presented to our hospital with the complaint of non healing episiotomy wound. She delivered a full term baby boy one and a half month back. Medio-lateral episiotomy was given during labour. Continuous suturing was done later with vicryl no 1. All the vital parameters were within the normal limits in the post operative period. Patient was discharged three days after the delivery. She visited the hospital for the follow up after three weeks. Episiotomy wound did not heal in this course of time and the patient also complained of diarrhea from past four days. Patient was prescribed tab augmentin, tab chymoral forte, cap ecoflora and high protein diet. Two weeks later patient presented with mild fever since two days. On examination, she had mild pallor. Episiotomy wound did not heal even after 5 weeks and gaping was noted (Figure1). No active bleeding was observed on per vaginal examination. A small mass was noted on per speculum examination. Laboratory investigations revealed haemoglobin 8.5gm/dl, mildly raised total leucocyte count and raised ESR. Patient was again admitted to the hospital in the view of above-mentioned findings. Excision of the mass was done under spinal

anaesthesia. Skin edges of the wound were cut, base of the wound and side walls were scraped. Skin edges were undermined with sharp scissors. Continuous suturing was done with vicryl no1. Tension suturing was done with prolene no 1. Patient was stable after the procedure and was fit for the discharge. Skin edges with the mass were sent for histopathology. Antibiotics with high protein diet and Sitz bath were prescribed. Histology report revealed a skin covered soft tissue mass measuring 3×2×1 cm. Cut surface was grey brown to grey yellow (Figure 2). Gross specimen was entirely sectioned and examined. Microscopically, the tissue lined by stratified squamous epithelium was seen. Focal areas show destruction of the epithelial lining and were replaced by the inflammatory cells. Acute and chronic inflammatory cells composed of mainly lymphocytes, few neutrophils and plasma cells admixed with xanthomatous cells were seen in the subepithelium (Figure 3a and 3b). Adipocytes were seen infiltrating upto the upper part of the sub epithelium (Figure 3a). There was no sign of granuloma, foreign body, and parasitic or fungal infection. The diagnosis of xantho-granulomatous inflammation with active chronic vaginitis was made.



Figure 1. Gaping of episiotomy wound.

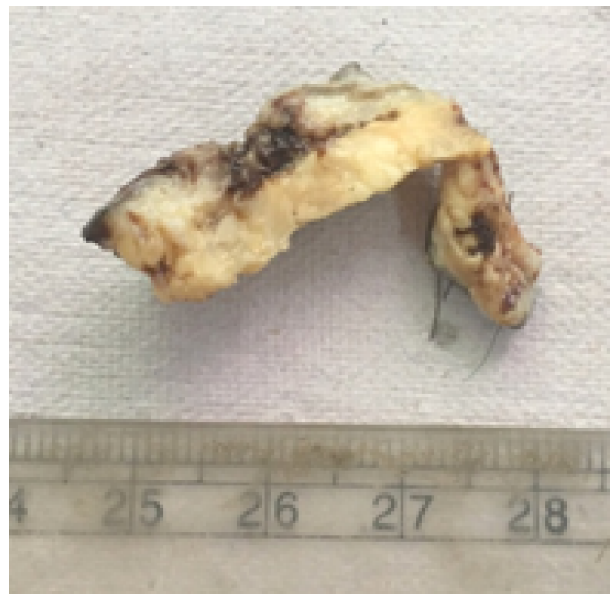


Figure 2. Skin covered soft tissue mass with grey white to yellow cut surface.

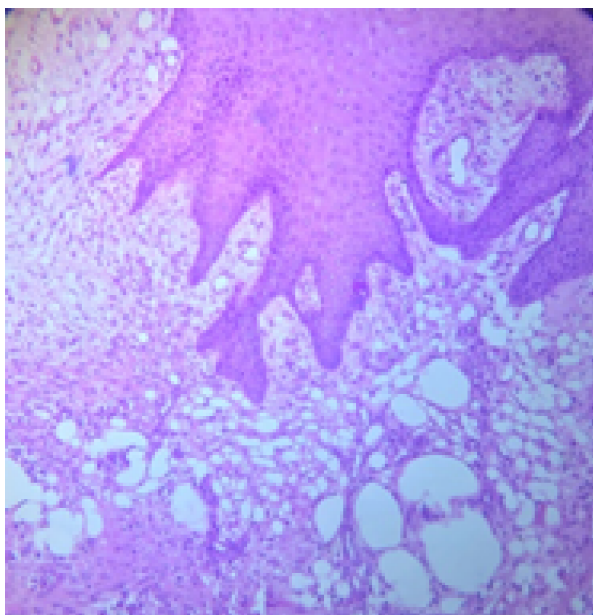


Figure 3a. Skin covered tissue with subepithelium showing inflammatory infiltrate admixed with xanthomatous cells (H&E stain, 4 X).

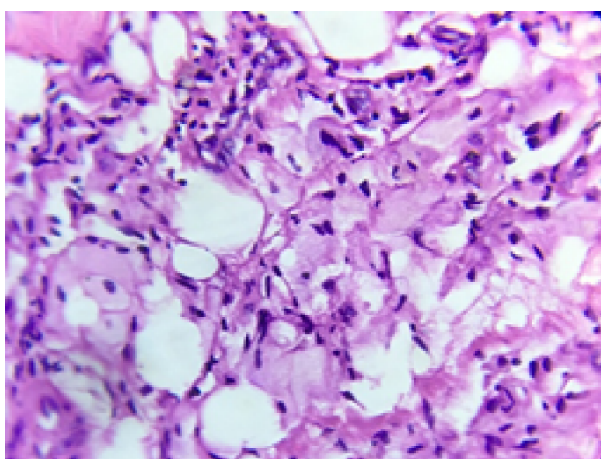


Figure 3b. Inflammatory infiltrate with xanthomatous cells (H&E stain, 40 X).

Discussion

Xanthogranulomatous inflammation (XGI) is a bizarre pathological identity characterized by the focal or diffuse destructive inflammatory process, with the accumulation of lipid-laden fibrous tissue, and acute and chronic inflammatory cells. It may involve any organ, but the most common sites are gallbladder and kidney^{1,2,3}. Incidence of XGI in the female genital tract (FGT) has not been established because of its rarity.

In 1976, Kunakemakorn et al. first described XGI of the FGT, which involves the uterine serosa, left fallopian tube and ovary⁴. It is rarely reported in vagina⁵. According to our search, XGI in the non healing episiotomy wound after normal vaginal delivery has not yet reported.

Various explanations were proposed for the mechanisms responsible for XGI, such as: immunological defect of the macrophage; chronic infection of the urachal diverticulum or cyst; foreign material such as retained suture material; abnormal lipid metabolism and lipid accumulation in a macrophage and gram negative or anaerobic bacteria such as in genitourinary tract infections or infection after tubal ligation⁶. Infections with *Bacteroides fragilis*, *Escherichia coli*, *Proteus vulgaris*, and *Salmonella typhi* were also considered. Among all these, the most accepted proposal was infection by *Escherichia coli* and other anaerobic bacteria. This was supported by clinical evidence of infection and growth of the bacteria from the affected tissue by culture⁷. Co-existent factors like bleeding and obstruction may also predispose to this infection. Tissue necrosis can occur as a result of all this, which is followed by the release of cholesterol and other lipids and phagocytosis by the macrophages⁶. Various etiological factors were suggested by Pang SY et al. in a literature review: pelvic inflammatory disease, intra-uterine contraceptive device, ineffective antibiotic therapy, endometriosis, leiomyoma, pelvic irradiation, inborn error in lipid metabolism, previous abdominal surgery, diabetes mellitus type 2, hyperlipidemia and untreated urinary infections⁸.

Majority of the cases of XGI of the FGT belongs to age range 23-72 years (mean age 32 years)⁷ and present with the complaints of fever, raised ESR and total leucocyte count. A well-defined mass on ultrasound is seen in most of the cases of XGI, which can be confused with the malignancy. Our patient presented with the complaints of gaping of non-healing episiotomy wound and fever. On examination, a small mass was felt, which was excised and sent for histopathology. Patient was put on antibiotics and was symptom free two weeks after the procedure.

Based on the clinical presentation and associated findings, the lesion is frequently mistaken for a malignant process⁹. But on histology XGI has a very clear-cut picture. Xanthoma cells (foamy macrophages) with abundant lipid laden cytoplasm and chronic inflammatory cells are consistently noted. Aggregates

of such foam cells are responsible for the yellow color observed on gross examination. The emergence of foam cells may be attributed to the following factors:

1. Inefficient or inappropriate antibiotics applied in the early phase of infection which resulted in ineffective control of bacterial multiplication. We also suspect the same to be the probable mechanism in our case.
2. Presence of a lipid metabolic disorder.
3. The application of intrauterine contraceptive devices or drugs resulting in adhesions with adjacent organs and pelvic peritoneum¹⁰.

Few inflammatory conditions such as malakoplakia, tuberculosis, actinomycosis and fungal infections may mimic XGI. The infections can be ruled out by culture and special stain for the causative organism and melakoplakia by the presence of intracytoplasmic concentric calcified bodies (Michaelis-Gutmann bodies)⁷.

Conclusion

XGI should be considered as one of the factors responsible for the delayed healing of the episiotomy wound. To the best of our knowledge, this is the first such case report depicting XGI in the non-healing episiotomy wound. Infection followed by the ineffective antibiotic therapy and the retained suture material may be the possible etiology in our case as the patient did not have any history of a lipid metabolic disease, nor did her history gave an indication of immunodeficiency.

Authors' contributions

All the authors contributed to the diagnosis and management of the patient. RN prepared the manuscript. All authors have critically revised and approved the final version of the manuscript.

References

1. Zhang XS, Dong HY, Zhang LL, Desouki MM, Zhao C. Xanthogranulomatous inflammation of the female genital tract: report of three cases. *J Cancer* 2012; 3: 100-6.
2. Pandey D, Saxena A, Rao AC, Valiathan M, Thomas J, Shetty J. Xanthogranulomatous inflammation in female pelvis: A diagnostic and therapeutic challenge. *Sri Lanka J Obstet Gynaecol.* 2015; 37: 18-20.
3. Gray Y, Libbey NP. Xanthogranulomatous salpingitis and oophoritis: a case report and review of the literature. *Arch Pathol Lab Med.* 2001 Feb;125(2):260-3.
4. Kunakemakorn P, Ontai H, Balin H. Pelvic inflammatory pseudotumor: a case report. *Am J Obstet Gynecol* 1976; 26: 286-7.
5. Narayan R, Mishra M, Mohanty R, Sinha TB. Xanthogranulomatous cervicitis. *Indian J Pathol Microbiol.* 2008; 51(3): 458-9.
6. Tai S, Liang C. A Case of Genitourinary Xanthogranulomatous Inflammation. *Gynecol Obstet* 2013; 3: 160.
7. Kumar SK, Sinha R, Kumar K and Shuchismita. Bilateral xanthogranulomatous oophoritis and salpingitis: a rare case report. *European J Pharmaceutical Medical Research* 2017; 4(5): 531-3.
8. Pang SY, Aggarwal IM, Lim YK. Xanthogranulomatous salpingo-oophoritis mimicking an ovarian malignancy – a series of 3 cases and review of literature. *Obstet Gynecol Int J* 2016; 5(3): 00162.
9. Russack V, Lammers RJ. Xanthogranulomatous endometritis: report of six cases and a proposed mechanism of development. *Arch Pathol Lab Med* 1990; 114: 929-32.
10. Bindu SM, Mahajan MS. Xanthogranulomatous oophoritis: A case report with review of literature. *Int J Health Allied Sci* 2014; 3: 187-9.