Pyometra Associated with Xanthogranulomatous Endometritis (XGE): A Case Report

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Abstract
Xanthogranulomatous endometritis (XGE) is a rare disease clinically simulating endometrial carcinoma [1,2]. It is a type of chronic inflammation comprising of lipid-laden macrophages (MØ), lymphocytes and multinucleated foreign body giant cells. Current lesion rarely develops in female genital system. So far 20 cases were reported worldwide[3]. Exact etiopathogenesis of the lesion is unknown. Several different bacteria e.g. Peptostreptococcus magnus, Escherichia coli, Klebsiella pneumoniae and Proteus vulgaris had been isolated earlier from similar lesion[4,5]. Present case presented with extensive collection of pus in uterine cavity. However, in the current case, pus culture was sterile and we could not isolate any microbe from pus.

Keywords: Pyogenic infection, foamy macrophages, chronic inflammation, lymphocytes.

Case Report
Present case was related to a 45-year-old female. She was operated and hysterectomy was done. Uterus was divided into two halves, measuring 5x3x2 cms and 3x2x1cms. Mucosal surface showed granular appearance (Figure 1a). Sections were taken from the endometrium. Section showed granulation tissue comprising of large number of lymphocytes and few plasma cells. Several multinucleated lipid-laden MØ were also seen (Figure 1b). Fibromuscular tissue of cervix showed dense lymphocytic infiltration, suggesting chronic cervicitis.

Xanthogranulomatous endometritis (XGE) is a rare disease, characterized by pain in lower abdomen, mild fever and foul-smelling pus discharge. About 40ml of pus was collected. Pus was cultured on blood agar and MacConkey agar medium; no organism was isolated. Hematological investigations revealed Hb 11.8gm/dl. Total leucocytes count was 12900 cells/mm³. Neutrophils were 80%, lymphocytes were 17% and monocytes were 3%. Radiologically, it showed loss of fat planes between uterus and posterior wall of urinary bladder and anterior wall of the rectum. Pus smear was stained by ZN method and examined; smear was negative for acid-fast-bacilli. Gram stained smear also did not show bacteria.
Figure 1 (a) shows gross granular mucosal endometrial surface. (b) Photomicrograph shows multiple giant cells (HEx 400). (c) Photomicrograph shows lymphocytic aggregate (HEx 600). (d) Photomicrograph shows foamy macrophages and lymphocytes (HEx 100). (e) Photomicrograph shows foamy macrophages (HEx 400). (f) Photomicrograph shows a multinucleated foreign body giant cell. Thick arrow denotes giant cell. Thin arrow denotes lymphocytes. Circle

Discussion

Exact etiopathogenesis of Xanthogranulomatous endometritis (XGE) is not known. Rarely, the disease may be fatal\[^4\]. Rarely, *Peptostreptococcus magnus*\[^2\], *Escherichia coli*, *Klebsiella pneumoniae* and *Proteus vulgaris*\[^5\] had been isolated. However, in the current case and in few other cases, no microbe was isolated\[^6,7\]. Another feature of the current case was thickened edematous endometrium due to chronic persistent inflammation. Microscopically, sections showed large number of lipid-laden MØ and multinucleate foreign body giant cells, suggesting granulomatous inflammation. Another feature of the current case was angiogenesis, suggesting granulation tissue formation. In addition, lymphoid tissue aggregates were seen. Further, small number of plasma cells were seen. Presence of lymphocytes and plasma cells was consistent with the diagnosis of chronic xanthogranulomatous inflammation. Recently, similar lesion involving myometrium had been reported and *Klebsiella pneumoniae* was isolated\[^8\].

*Klebsiella pneumoniae* is an encapsulated bacteria. Capsule contains a neutral polypeptide which inhibits neutrophils\[^9\]. Therefore, neutrophils were not seen in the exudate of present case. Other microbes e.g. *Peptostreptococcus magnus*, *Proteus vulgaris*, *Klebsiella pneumoniae* and *Escherichia coli* might have a role in pathogenesis of XGE. *Mycoplasma* had been speculated to cause XGE. *Mycoplasma* remains outside the endometrial cells and cause injury and death of endometrial cells\[^9\].

Conclusion

Exact etiopathogenesis of XGE is not known. Several bacteria have been reported e.g. *Peptostreptococcus magnus*, *E. coli* and *Proteus vulgaris* had been isolated from several cases. However, in the current case no microorganism
could be isolated. In the current case, endometrium showed features of granulomatous inflammation. Presence of giant cells suggested possible role of mycobacteria in pathogenesis of XGE. Anti tuberculous treatment had been tried in a few cases without much relief.

References