Case Report
Obstet Gynecol Sci 2016;59(5):411-414
http://dx.doi.org/10.5468/ogs.2016.59.5.411
pISSN 2287-8572 · eISSN 2287-8580

Obstetrics & Gynecology Science

Cervical xanthogranuloma in a case of postmenopausal pyometra

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Xanthogranuloma is a non-neoplastic presentation of chronic inflammation commonly seen in gallbladder, kidney and rarely seen in genital organs. Only one case has been reported in cervix. Here, we report a case of 60-year-old postmenopausal lady who presented with history of fever and purulent discharge per-vaginum. On speculum examination, cervix had an ulcer extending from 3 to 5 o'clock position. Uterus was bulky. On probing the ulcer, a 1-cm deep sinus was identified. Ultrasound showed enlarged uterus and fluid collection suggestive of pyometra. Pyometra was drained and cervical biopsy was taken from the ulcerated lesion; histopathology revealed granulomatous inflammation with predominantly xanthous cells suggestive of tuberculosis. High index of clinical suspicion needs to be maintained in abnormal cervix. It is a perplexing and rare entity for a clinician and also a diagnosis of exclusion; only histopathology can help for diagnosis. It mimics like malignancy and chronic infections.

Keywords: Cervix uteri; Tuberculosis; Xanthogranulomatous inflammation

Introduction

Xanthogranulomatous inflammation (XGI) is an uncommon form of chronic inflammation that is histopathologically characterized by a marked proliferative fibrosis, parenchymal destruction, and infiltration of foamy histiocytes intermixed with hemosiderin laden macrophages and foreign body giant cells. It has been reported in multiple organs, most commonly in the kidney, gall bladder, salivary glands and bones and less commonly in the female genital tract [1-4]. In genital organ it mostly affects the endometrium, but involvement of the fallopian tube, ovary, cervix and vagina may also rarely occur [4]. Narayan et al. in 2008 [5] has reported the first case of xanthogranuloma of cervix.

Case report

A 60-year-old P4L4 postmenopausal woman presented with chief complaints of foul smelling discharge per vaginum with low grade fever on and off since 1 year after that she had high grade fever since 1.5 months. She had menopause at 50 years and had never taken hormone replacement therapy. There was strong family history of tuberculosis in her close relatives. Her general physical examination was unremarkable. Her per-speculum examination showed unhealthy cervix, ulcerative lesion on the posterior lip of cervix from 3 to 5 o'clock position and pus was coming freely from that area (Fig. 1A). On probing it, that area was 1 cm in depth. On pelvic examination uterus was bulky in size, bilateral fornices free and non-tender. In her complete blood picture, total leucocytes count was 10,000 cells/μL, with neutrophils 63% and lymphocytes 32%. Her erythrocyte sedimentation rate was raised (65 mm) and montoux test was positive. Her Pap smear was unsatisfactory due to low cellularity.

Ultrasound revealed a fluid collection in uterine cavity around 70 ml. In contrast-enhanced computed tomography

Received: 2015.11.10. Revised: 2016.2.17. Accepted: 2016.3.2.
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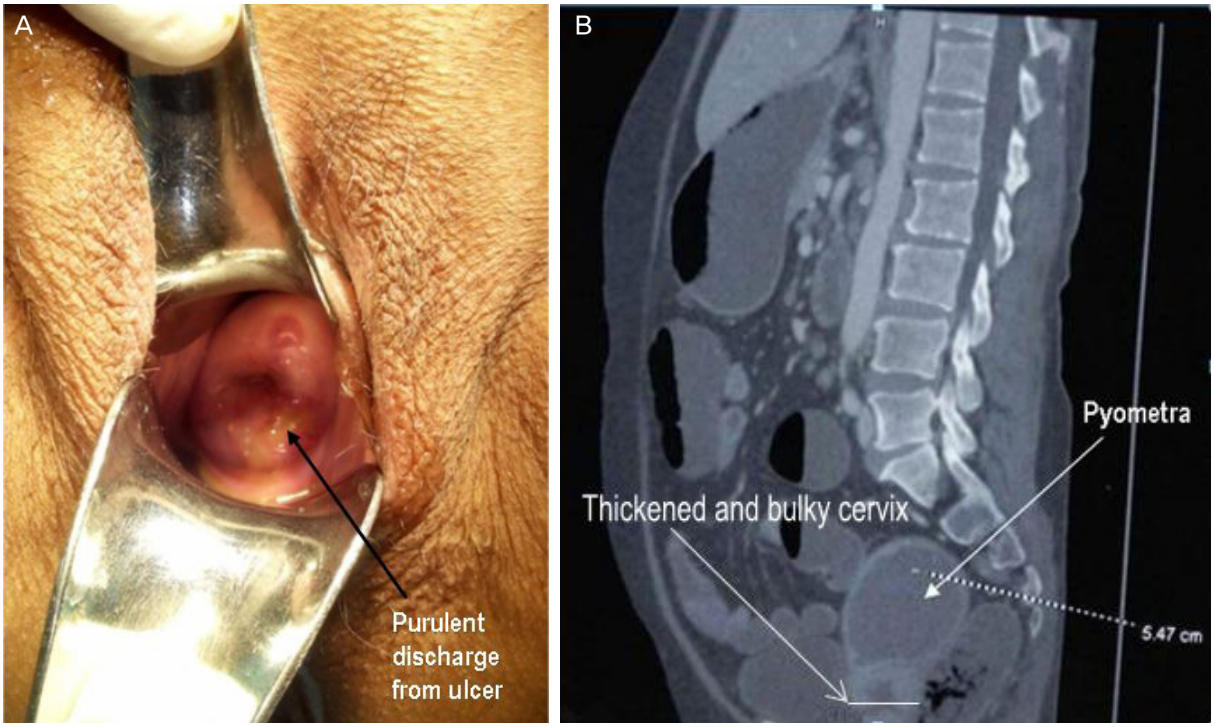

Fig. 1. (A) Ulcerative lesion on the posterior lip of cervix from 3 to 5 o'clock showing purulent discharge. (B) Contrast-enhanced computed tomography pelvis showing pyometra, obliteration of fat planes between uterus and bladder; uterus and rectum.

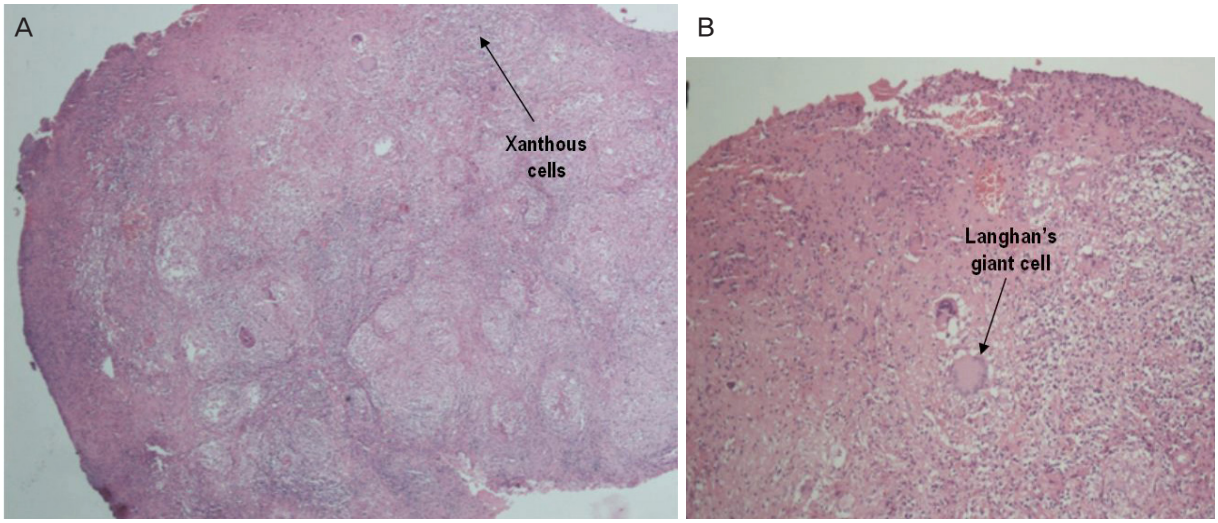

Fig. 2. (A) Histopathology slide of cervical biopsy showing numerous granulomas in the cervical stoma (H&E, ×40). (B) Epithelioid cell granuloma with Langhan's giant cell is seen beneath the ulcerated epithelium (H&E, ×100).

cervix was bulky, heterogeneously enhancing thickened both lips of cervix, and endometrial cavity was distended with fluid suggestive of pyometra, obliteration of fat planes between uterus and bladder; uterus and rectum (Fig. 1B). Also there were multiple enlarged lymph nodes like portocaval, retrocaval, precaval, aortocaval, preaortic and para-aortic in contrast-enhanced computed tomography of abdoman and pelvis. All these findings can occur in malignancy as well as inflammation.

Patient has undergone pyometra drainage and cervical biopsy from posterior lip of cervix was done. Her peroperative finding was 50 mL of pus drained which was thick and

cheesy. Her symptoms persists inspite of empirical antibiotic therapy given for 2 weeks. Final diagnosis was made only after histopathological examination of cervical biopsy which showed XGI of cervix likely of tubercular etiology. Numerous granulomas are seen the cervical stroma. The surface epithelium is ulcerated at most places (H&E, ×40) (Fig. 2A). Epithelioid cell granuloma with Langhan's giant cell is seen beneath the ulcerated epithelium (H&E, ×100) (Fig. 2B).

Although her smear for acid fast bacilli was negative and BACTEC with radiometric culture (the radiometric method; Becton Dickinson, Towson, MD, USA) was negative, patient was started on antituberculosis treatment (ATT) category 3 regimen (2 months of isoniazid, rifampicin, and pyrazinamide; 4 months of isoniazid and rifampcin) because of raised erythrocyte sedimentation rate, Mantoux positive, strong family history of tuberculosis and histopathology report. Although endometrial sampling was not taken and planned at second sitting but she improved gradually on ATT and hence endometrial sampling deferred later on also. She has been followed for 6 month and became asymptomatic.

Discussion

XGI is chronic inflammatory disorder which affect multiple organs of body usually involve kidney, gall bladder, salivary gland and bones, involvement of female genital tract is rare and mostly affect endometrium, rarely involve vagina, cervix, ovary, fallopian tube. The exact etiology of the disease is not known; however most likely etiopathogenesis are suppurative infections, organ obstruction, and hemorrhage trigger leads to tissue damage within the involved organs, usually eliciting a microscopic response of the disease process. XGI causes destruction of the involved organ and could be misinterpreted as a locally invasive cancerous lesion. Biedermann et al. [6] supports the infective etiology theory of this condition following long-standing infections with mycoplasma hominis in a patient of tubo-ovarian abscess leading to peritonitis. Similarly, in our case, culture of purulent vaginal discharge showed *Escherichia coli*. On extensive search of literature, with key words cervical xanthogranuloma, xanthogranuloma of female reproductive tract, genital tuberculosis, till date total 20 cases of XGI of endometrium and 16 cases of XGI of fallopian tubes and ovary [7-15] has been reported.

The first case of xanthogranulomatous endometritis was described by Barua et al. in 1978 [7] and last by Wader et al. in 2013 [8] mimicking carcinoma cervix. Two cases were associated with haematometra, two with carcinoma with endometrium, one infection with enterococcus species and Proteus magnus, in others cause not known. Narayan et al. [5] reported first case of xanthogranuloma of cervix in 2008 mimicking carcinoma cervix and our may be the second.

Xanthogranuloma cervix is a rare entity, difficult to diagnose clinically and for a clinician it is diagnosis of exclusion. It is basically histopathological diagnosis which is different from other chronic inflammatory conditions like tuberculosis. As in our case, inspite of specific antibiotic (tazobactam + piperacillin and amikacin) given as per culture sensitivity; her condition has not improved. But after histopthological report, cervical XGI likely due to tuberculosis, even BACTEC culture was negative, she has responded to ATT. So, our hypothesis also supports infective etiology which can mimic like malignancy. Biedermann et al. [6] has also found congolomere tumour due to inflammation in the case of tuboovarian mass; similarly in our case computed tomography abdomen and pelvis findings revealed obliteration of fat planes between uterus and bladder; uterus and rectum which may be because of malignancy or inflammation. Tuberculosis was not associated with any of the above mentioned case reports. In some cases in which diagnosis has already been made by biopsy, antibiotic therapy with doxycycline etc [6], removal of source of inflammation like spillage of gallbladder content, removal of foreign implant in case of xanthogranuloma breast resulting from breast implants etc, were done with consequent improvement of the clinical condition of the patient and the resolution of inflammatory markers. In some cases diagnosis was made after surgery like salphingo opherectomy and hysterectomy and in some antibiotics tried but failed, and surgery was needed [13,14]. This is the first case XGI which was treated by ATT even though tuberculosis was not associated with any of the previous cases of XGI.

Tanwar et al. in 2015 [14] reported xanthogranulomatous salpingo-oophoritis in youngest girl of 2 years. So, XGI can occur at any age group, in our case it was in 70 years lady. XGI with infiltration into the uterine myometrium from the parametrium without endometritis was reported by Inoue et al. in 2014 [15].

Cervical XGI is very rare entity and it mimic like malignancy clinically as well as radiologically. Pathologist should be very prompt in diagnosis of such rare diseases. Though this case

was diagnosed as XGI but pathologist also commented it likely of tubercular in origin. By seeing her clinical profile and strong family history of tuberculosis, antitubercular drugs were started and her symptoms resolved. Through this case report we want to highlight in literature that unnecessary hysterectomies can be avoided by prompt diagnosis of rare entities.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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