Case Report

Malakoplakia of Endometrium with Osseous Metaplasia on Evaluation of Postmenopausal Leukorrhea: A Rare Case Report

Yavana Suriya Venkatesh, S Norton Stephen[1], Murali Subbaiah, Bhawana A. Badhe[1], Gowri Dorairajan

Departments of Obstetrics and Gynaecology and [1]Pathology, Jawaharlal Institute of Postgraduate Medical Education and Research, Puducherry, India

Abstract

Malakoplakia is a chronic xanthogranulomatous condition that affects the genitourinary tract reported earlier as urinary granulomas and pelvic masses. We report a different clinical manifestation of malakoplakia presenting as postmenopausal pyometra. A 64-year-old postmenopausal female presented with foul-smelling vaginal discharge with a past history of induced abortion, followed by dilatation and evacuation. On examination, abdomen was soft, vaginal examination revealed pus discharge, parous size uterus with free fornices, and pap smear ruled out malignancy. Ultrasonography revealed linear, echogenic structures in the endometrial cavity suspicious of bony spicules with fluid around. Hysteroscopy revealed congested endometrium with multiple pieces of shredded bone-like structures that were removed followed by curettage. Histopathological examination was suggestive of malakoplakia with osseous metaplasia. Retained bony spicules can cause chronic granulomatous inflammation that may become symptomatic postmenopause due to absent cyclical shedding. This is the first reported case of malakoplakia of uterus following retained bony spicules.

Keywords: *Case report, malakoplakia of uterus, MichaelisGutmann bodies, osseous metaplasia, pyometra, von Hansemann cells*

Submitted: 12-Sep-2020
Revised: 10-Aug-2021
Accepted: 28-Aug-2021
Published: 16-Oct-2021

Introduction

Malakoplakia is a chronic granulomatous disease of infectious etiology that usually involves the urinary bladder, testicles, prostate, lymph node, and skin in immunocompromised individuals. Uterine malakoplakia is very scarcely reported in the literature. Endometrial osseous metaplasia is another rare and distinct entity referring to the presence of bone-like tissue in the uterine cavity. Both the conditions have varied clinical presentation, risk factors, and controversial etiopathogenesis. We report a case of uterine endometrial malakoplakia coexistent with osseous metaplasia.

Case Report

A 64-year-old postmenopausal female, P5 L5A1, attended a gynecological clinic with a history of mucoid to foul-smelling white discharge per vagina for the past 6 months. Her previous menstrual cycles were regular, the perimenopausal transition was smooth, and she attained menopause 14 years ago. She was previously asymptomatic with no history of postmenopausal bleeding. She had five spontaneous vaginal deliveries and one termination of pregnancy (medical termination of pregnancy [MTP]). Her antepartum and postpartum periods were uneventful with five living issues and her last childbirth 35 years back. Thirty years back, she underwent MTP at 16 weeks by dilatation, evacuation, and history of excessive bleeding, requiring blood transfusion. No other history of past surgeries or tuberculosis. She is a well-controlled hypertensive on medication and coronary artery disease, for which she underwent percutaneous transluminal coronary angioplasty and stenting 5 years ago.

On examination, general physical examination was unremarkable. The abdomen was soft, with no mass or

Access this article online

Quick Response Code:

Website: www.jmidlifehealth.org

DOI: 10.4103/jmh.jmh_213_20

Address for correspondence: Dr. S Norton Stephen, Department of Pathology, Jawaharlal Institute of Postgraduate Medical Education and Research, Puducherry, India.
E-mail: norton.stephen89@gmail.com

How to cite this article: Venkatesh YS, Stephen SN, Subbaiah M, Badhe BA, Dorairajan G. Malakoplakia of endometrium with osseous metaplasia on evaluation of postmenopausal leukorrhea: A rare case report. J Mid-life Health 2021;12:237-40.

organomegaly. On gynecological examination, the vulva and vagina were atrophic. Per speculum revealed pus-like discharge with normal ectocervix. The uterus was enlarged to parous size, mobile with free fornices. Pap smear was inflammatory and negative for intraepithelial lesion or malignancy. Her ultrasonography revealed an enlarged uterus of 10.7 cm × 7.5 cm filled with linear, echogenic structure in the endometrial cavity up to the cervix with fluid around. The endometrial cavity showed evidence of pyometra. Differential diagnoses on ultrasound were suspected retained bones or dystrophic calcification. Magnetic resonance imaging done showed evidence of pyometra of 39 mm × 26 mm × 31 mm and bone-like echogenicity in the endometrial cavity. The patient underwent hysteroscopy, which revealed multiple pieces of shredded bony structures embedded in the endometrial cavity, all removed using forceps [Figure 1]. The endometrial cavity did not have any abnormal vascularity or hyperplastic changes. Endometrial sampling was done, and specimens were sent for histopathological examination.

The microscopic examination of the paraffin-embedded H and E sections showed woven bone in bony bits, and endometrial tissue showed foamy histiocytes with multiple tiny calcareous round basophilic bodies (Michaelis-Gutmann bodies) along with lymphocytes, hemorrhage, and fibrin [Figure 2]. Von Kossa and periodic acid Schiff (PAS) stain highlighted the basophilic bodies; Perl's stain highlighted the iron pigment [Figure 3]. Histopathological diagnosis of malakoplakia with osseous metaplasia was made.

Discussion

Malakoplakia (malakos-soft, plakos-plaque) was first described in the year 1901 by von Hansemann and in 1902 by Michaelis and Gutmann. Uterine malakoplakia is rarely reported in the literature and has a broad clinical spectrum and findings. Few cases of malakoplakia have been reported in the literature presenting as either cervical ulcerative polyp,[1] as pelvic mass with extensive malakoplakia involving ovary, uterus, and fallopian tube[2] and as endometrial malakoplakia on the evaluation of postmenopausal bleeding.[3]

Our patient presented with chronic postmenopausal white discharge per vagina with suspected minimal pyometra. Malignancy causing pyometra is always the highest suspicion in postmenopausal women with pus discharge. In our case, the sonographic picture of intense linear echoes with shadowing made us suspicious of bones likely retained during the past abortion. We proceeded with a hysteroscopy. Surprisingly, the patient became symptomatic after so many years. The previous

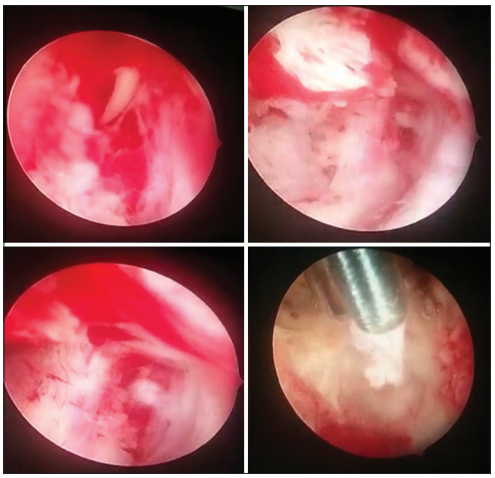

Figure 1: Images of hysteroscopy showing multiple bone-like structures in the endometrial cavity

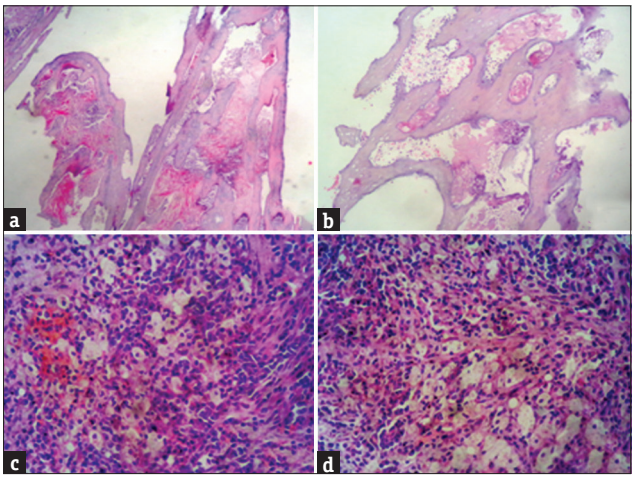

Figure 2: H and E sections showing woven bone in bony bits (a and b) and endometrial tissue showed foamy histiocytes with multiple tiny calcareous round basophilic bodies (Michaelis-Gutmann bodies) along with lymphocytes, hemorrhage, and fibrin (c and d) -1800 x 1200 pixels

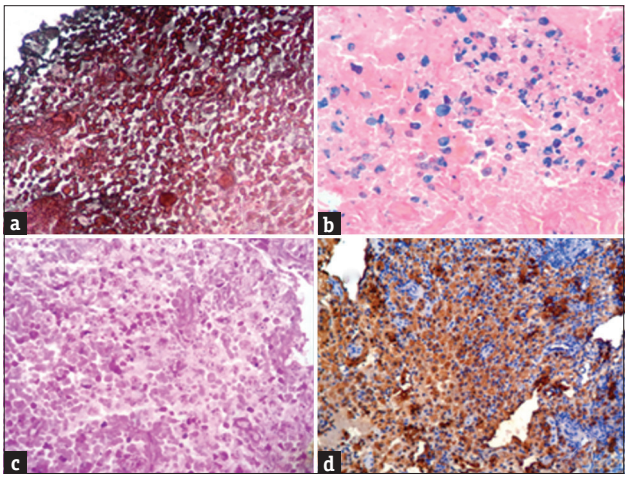

Figure 3: von Kossa (a) and periodic acid Schiff stain (b and c) highlighting the basophilic bodies, Perl's stain highlighting the iron pigment -1800 x 1200 pixels, 1.(d) CD 68 antibody [Immunohistochemical stain] highlighting the histiocytes

menstrual cycles were regular even after second-trimester dilatation and evacuation with no postmenopausal

bleeding. It is possible that atrophic endometrium loses the defense mechanism and results in late symptoms.[4]

The pathophysiology underlying malakoplakia is the defective phagolysosomal activity, leading to the accumulation of partially digested bacteria in macrophage with calcium and iron deposition in the residual bacterial glycolipid. The organisms associated include *Escherichia coli*, *Klebsiella*, *Proteus*, and *Mycobacterium tuberculosis*.

The histopathological diagnosis is made by the presence of von Hansemann cells that are enlarged macrophages with foamy cytoplasm, eccentric hyperchromatic nuclei with intracytoplasmic, and extra cytoplasmic inclusion bodies (von Kossa positivity due to calcium carbonate deposition and PAS stain positivity). The bacterial degradation products provide a matrix for deposition of needle-like crystals resembling hydroxyapatite that are round to oval concentric, laminated inclusions termed Michaelis-Gutmann bodies.[5]

Endometrial osseous metaplasia, a coexistent finding, in this case, is another rare entity with the reported incidence of 3 in 10,000.[6] The proposed etiologies include heterotopia, ossification of postabortive endometritis,[7] retained fetal bones, endometrial tuberculosis, and dystrophic calcification,[5] with a varied clinical presentation such as pelvic pain menstrual abnormalities, infertility, and white discharge. Osseous metaplasia is known to alter the milieu of the endometrium by increasing the production of prostaglandins. This mechanism explains the presentation in our patient wherein the previous second-trimester termination by dilatation and evacuation would have possibly led to chronic endometritis and metaplastic endometrial osseous metaplasia and Gram-negative bacterial infection secondarily leading to malakoplakia. The medical methods of second-trimester abortions adopted nowadays are safer and cause reduced endometritis compared to the previously practiced methods of aspirotomy.

Two hypotheses support the clinical presentation in the index patient. One is the osseous metaplasia secondary to chronic endometritis, subsequently leading to malakoplakia. Another possibility is the retained fetal bones[8] embedded in the endomyometrium, which probably might not have got dislodged during menstruation, similar to a retained neglected intrauterine contraceptive device. The chronic inflammation may have led to the development of uterine malakoplakia, explaining the patient's symptomatology. The time interval between antecedent abortion and the detection of endometrial ossification has been reported in the literature to range from 8 weeks to 15 years, explaining the chronicity of the condition.[9]

Management options include either medical or surgical depending on the extent and associated sites of involvement. Antibiotics such as aminoglycosides, quinolones, and cotrimoxazole[10,11] have been tried with or without surgery in the treatment of malakoplakia. Retrieval of osseous fragments and curettage has been advocated for uterine osseous metaplasia.[12] Hysteroscopic removal of bone-like structures, curettage, and treatment with antibiotics (ciprofloxacin and metronidazole) was therapeutic in our patient, and she is asymptomatic at 1-year follow-up now. It is necessary to do hysteroscopy as in the index case for the complete retrieval of suspected bony spicules and biopsy from suspicious areas after the direct visualization of the cavity. Blind dilatation and curettage alone would be an incompetent way and hence not advisable for either diagnosis or treatment.

The methods to prevent such pathological conditions include the practice of safer abortion methods as per the guidelines, ensuring completeness of the procedure when advocated and follow-up care. Detailed clinical history, thorough examination, ultrasound imaging, Doppler for vascularity, and hysteroscopy as indicated in abnormal bleeding or leukorrhea would clinch the diagnosis earlier in such patients, and prompt management will avoid chronicity of the condition. Endometrial osseous metaplasia with malakoplakia mimics malignancy, but they are benign pathological conditions wherein the diagnosis will alter the management line to a conservative approach. The risk of malignant transformation is not well studied due to the rarity of the condition perse, but there is documented evidence of malignancies coexisting with malakoplakia, which needs to be ruled out.[13] This case report provides insight into the varied clinical presentation, diagnosis, and successful management of uterine malakoplakia and aids the treating gynecologist during subsequent workup and the evaluation of persistent leukorrhea.

Conclusion

Although a rare entity, malakoplakia of the endometrium and uterine osseous metaplasia may be considered in the differential diagnosis of postmenopausal bleeding or leukorrhea when the diagnosis is uncertain, prevention of the disease by adopting safe abortion methods, early diagnosis, ruling out malignancy, recognition, and apt medical/surgical management is advocated.

Informed consent

We have obtained informed consent from the patient. The institution of study (Jawaharlal Institute of Post

Graduate Medical Education and Research, Puducherry) gives approval on informed consent from the patient for case reports. The institute does not mandate a separate ethical approval for reporting cases.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that their name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Eckford SD, Hirschowitz L, Brown I, Akande V. Malakoplakia of the uterine cervix – A rare cause of postmenopausal bleeding. J Obstet Gynaecol 1994;14:51-2.
2. Chou SC, Wang JS, Tseng HH. Malacoplakia of the ovary, fallopian tube and uterus: A case associated with diabetes mellitus. Pathol Int 2002;52:789-93.
3. Ehara T, Korematsu M, Kawamura K, Shimoji K, Matsui H, Shimizu K. Malakoplakia of the endometrium. J Jpn Soc Clin Cytol 2013;52:139-42.
4. Otify M, Fuller J, Ross J, Shaikh H, Johns J. Endometrial pathology in the postmenopausal woman – An evidence based approach to management. Obstet Gynaecol 2015;17:29-38. Afonso JP, Ando PN, Padilha MH, Michalany NS, Porro AM. Cutaneous malakoplakia: Case report and review. An Bras Dermatol 2013;88:432-7.
5. Singh P, Kapur K, Singla S, Naz N. Endometrial osseous metaplasia and mature bone formation with extramedullary hematopoiesis. J Hum Reprod Sci 2011;4:56-7.
6. Bahçeci M, Demirel LC. Osseous metaplasia of the endometrium: A rare cause of infertility and its hysteroscopic management. Hum Reprod 1996;11:2537-9.
7. Umashankar T, Patted S, Handigund R. Endometrial osseous metaplasia: Clinicopathological study of a case and literature review. J Hum Reprod Sci 2010;3:102-4.
8. Cayuela E, Perez-Medina T, Vilanova J, Alejo M, Cañadas P. True osseous metaplasia of the endometrium: The bone is not from a fetus. Fertil Steril 2009;91:1293.e1-4.
9. Christensen M, Knuhtsen S, Knudsen E. Colonic malacoplakia treated with sulfamethizole and trimethoprim. Ugeskr Laeger 2011;173:509-10.
10. Dong H, Dawes S, Philip J, Chaudhri S, Subramonian K. Malakoplakia of the urogenital tract. Urol Case Rep 2015;3:6-8.
11. Coccia ME, Becattini C, Bracco GL, Scarselli G. Ultrasound-guided hysteroscopic management of endometrial osseous metaplasia. Ultrasound Obstet Gynecol 1996;8:134-6.
12. Pillay K, Chetty R. Malakoplakia in association with colorectal carcinoma: A series of four cases. Pathology 2002;34:332-5.
13. Darvishian F, Teichberg S, Meyersfield S, Urmacher CD. Concurrent malakoplakia and papillary urothelial carcinoma of the urinary bladder. Ann Clin Lab Sci 2001;31:147-50.