Case Report

DOI: 10.21276/APALM.3151

Osteocartilaginous metaplasia in the Endometrium: A Rare Reversible Cause of Secondary Infertility

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ABSTRACT

Osteo-cartilaginous metaplasia in endometrium is an infrequent finding and usually presents with secondary infertility. This is a benign entity and usually associated with a previous history of abortion. It can be treated successfully with hysteroscopy and henceforth reverting infertility. Awareness of this entity is important in order to avoid overdiagnosis of mixed Mullerian tumor of the endometrium followed by hysterectomy. Herein, we present one such case of a 28-year-old female who presented with secondary infertility.

Keywords: *Endometrial osteo- cartilaginous metaplasia, malignant mixed müllerian tumor*

Introduction

Metaplasia is defined as the transformation of one differentiated cell type to another cell type, either homologous or heterologous, which may be part of a normal maturation process or caused by some sort of abnormal stimulus. Mesenchymal metaplasia of endometrium is a rare entity, although epithelial metaplasia is more frequently encountered. Stromal metaplasia includes formation of islands of smooth muscle, cartilage, synovium, and bone. More than 100 cases have been reported in the world literature having either osseous or cartilaginous metaplasia in the endometrium but only one case report [1] documenting the combined osteo cartilaginous metaplasia has been identified till date. We put forward the second case having both the heterologous elements in the endometrium.

The mainstay of this case report is to create awareness regarding this treatable entity and avoid overdiagnosis of carcinosarcoma/malignant mixed müllerian tumor.

Case Report

A 28-year-old female patient presented to gynecology outpatient department with the history of secondary infertility, irregular bleeding p/v and pain in abdomen. She was married for 3 years and had a history of medical termination of pregnancy two years back. Her menstrual cycles were irregular since then and was associated with pain in abdomen.

She underwent ultrasonography which revealed thickened endometrium with foci of calcification at lower uterine segment. The patient was subjected for a diagnostic hysteroscopy and curetting. The sample was then sent for histopathological examination.

The provided curetted sample was routinely processed. The Hematoxylin & Eosin-stained sections showed viable endometrial tissue comprising of endometrial glands embedded in cellular stroma containing thick-walled blood vessels and infiltrated by lymphocytes, plasma cells and polymorphs. [Figure- 1] In addition, islands of mature bony trabeculae along with marrow elements and foci of cartilage were observed. [Figure- 2, 3] Necrosis, granulomatous pathology or any products of conception were not identified. The serum calcium and phosphorus were within normal limits. Compiling all the clinical, radiological, and histopathological findings, a final diagnosis of osteo-cartilaginous metaplasia of endometrium associated with chronic endometritis was made.

Discussion

Endometrial metaplasia refers to the replacement of the normal endometrial glandular epithelium or stroma by cells that are usually not encountered in the normal endometrium. Epithelial metaplasia, which is commonly seen include squamous, mucinous, tubal, eosinophilic, papillary, secretory and hobnail types. Whereas, stromal metaplasia, which is infrequent, includes osseous, cartilaginous, myomatous, adipose and synovial-like. [2] Because these cells are unusual at the site, this benign entity can be misinterpretated as malignancy. [1,3]

There are several theories proposed for such metaplastic transformation. Most common factor being the chronic inflammation, especially genital tuberculosis in India. [4] Others inflammatory factors include chronic endometritis, pyometra or inflammation following retained product of conception.[5] These chronic insults to the endometrium

stimulates the proliferation of multipotent cells that can differentiate into either chondroblasts or osteoblasts. [6]

Other proposed mechanisms include hypercalcemia, hyperoestrogenism, iatrogenic implantation of the fetal tissue, including the cartilage, into the uterine wall following dilatation and curettage. [7]

It has broad range of clinical presentation varying from menstrual irregularities, pelvic pain, dyspareunia, vaginal discharge to even secondary infertility. [8] Like the present case, many of the previous published cases had reported secondary infertility [9-12] and subsequent conception with conservative treatment approach. Hysteroscopic evacuation of bony and cartilaginous tissue has always been the treatment of choice. Earlier it was recommended to remove the bone from the endometrium by a series of dilatation and curettage to avoid synechiae formation. [11,12] But nowadays, recent studies recommend ultrasound assisted hysteroscopic removal of the bone and cartilage to aid proper visualization and thorough removal of tissue from endometrium as well as those embedded in the myometrium. [4, 7, 12, 13]

Knowing its non-neoplastic nature and a conservative approach of management, this condition needs to be recognized by pathologists to avoid making an overdiagnosis of malignant mixed mullerian tumor of the uterus. [7, 11, 13]

Conclusion

Endometrial osteo-cartilagous metaplasia is rare benign entity that is either overlooked or misdiagnosed as malignant mixed mullerian tumor of the uterus. It commonly presents as secondary infertility and is treatable with hysterocsopic evacuation of osteo-cartilagenous tissue. Hence, a careful histopathological examination is required to avoid unnecessary hysterectomy in young females.

Acknowledgements

We are thankful to Dr. Nachiketa Mohapatra and Dr Biswajit Mohanty for their constant academic support.

Funding

None.

Competing Interests

None declared.

Consent

A written informed consent has been taken from the patient for use his personal information in academic and publication work. The Ethical committee of our institution has approved this case report.

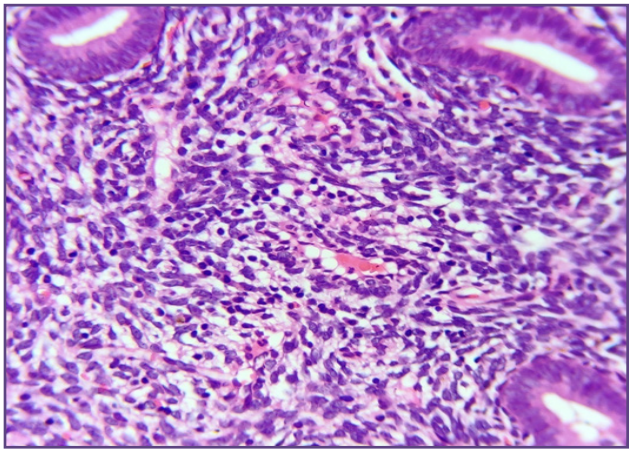

Fig. 1: Photomicrograph showing endometrial stroma infiltrated by lymphocytes, plasma cells (Arrow) and polymorphs (H&E; 400x).

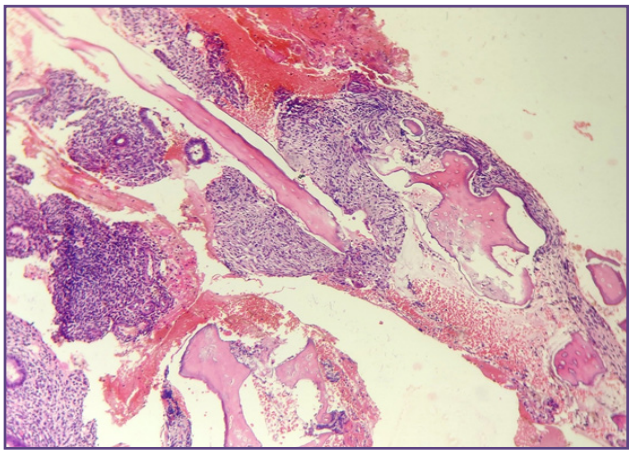

Fig. 2: Photomicrograph showing islands of mature bony trabeculae embedded in the endometrial stroma (H&E; 100x).

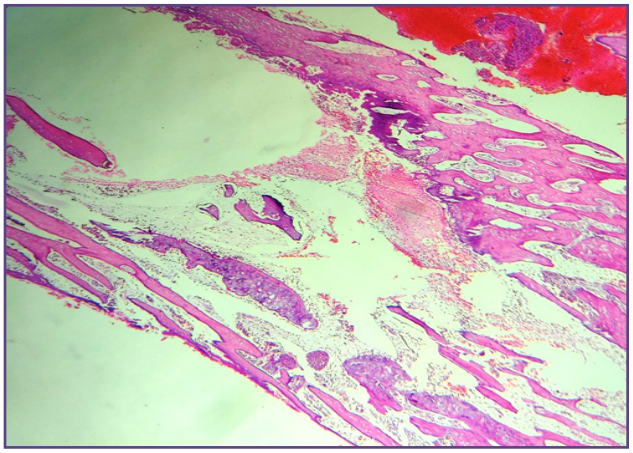

Fig. 3: Photomicrograph showing islands of mature bony trabeculae and cartilage tissue [Arrow] (H&E; 40x).

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Financial or other Competing Interests: None.

Date of Submission : 21/01/2022
Date of Final Revision : 20/03/2022
Date of Acceptance : 31/03/2022
Date of Publication : 30/04/2022