Case Report

Endometrial osteocartilaginous metaplasia: A case report and review of the literature

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Abstract

Rare cases of osseous metaplasia and a single case of cartilaginous metaplasia have been reported previously but no previously reported cases of osteo-cartilagenous metaplasia were found on searching the literature. Osteo-cartilagenous metaplasia is a benign lesion which may be treated successfully with hysteroscopy. Awareness of this condition by histopathologists is important in order to avoid misdiagnosis of endometrial malignant mixed mullerian tumour. The authors report a patient with endometrial osteo-cartilagenous metaplasia, describe the clinical and histological features and review the literature on this condition.

Keywords: Cartilaginous metaplasia, endometrium, osseous metaplasia, osteocartilaginous metaplasia

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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. In the endometrium, the metaplastic process can be seen in both the epithelial and rarely the mesenchymal components. The presence of endometrial metaplasia can significantly complicate the histological interpretation of endometrial biopsy material due to glandular architectural complexity, crowding, and presence of heterologous elements, which can lead to an erroneous diagnosis of endometrial hyperplasia or malignancy if a pathologist is unaware of the potential pitfalls.[1]

Access this article online	
Quick Response Code:	Website: www.ijcpc.org
	DOI: 10.4103/ijcpc.ijcpc_13_18

Osseous and cartilaginous metaplasias are both unusual forms of mesenchymal metaplasia in the endometrium.[2] Rare cases of osseous metaplasia and cartilaginous metaplasia have been reported previously; however, no previously reported cases of osteocartilaginous metaplasia were found on searching the literature. Histological samples taken at hysteroscopy would be expected to show foci of bone and cartilage admixed with endometrial tissue. The features may be erroneously misinterpreted as heterologous elements of a malignant mixed Mullerian tumor if the diagnosis of osteocartilaginous metaplasia is not considered.

CASE REPORT

A patient is a 27-year-old female who presented to reproductive medicine primary subfertility. Other significant medical history includes polycystic ovary syndrome and HIV. She is also a sickle cell carrier.

How to cite this article: Ching Y, Gudi M, Vella JE. Endometrial osteocartilaginous metaplasia: A case report and review of the literature. Int J Clinicopathol Correl 2018;2:34-6.

She underwent an ultrasound examination which showed a thickened endometrium with a calcified polyp [Figure 1]. Hysteroscopy and curettings were subsequently performed, and what was thought to be a calcified polyp was identified within the endometrial cavity and biopsied.

Histological examination showed blood admixed with fragmented pieces of endometrial tissue, showing stromal clumping and glandular disintegration in keeping with menstrual breakdown. In addition, islands of mature bone and cartilage were seen, suggestive of osteocartilaginous metaplasia. The surrounding viable endometrial tissue showed a plasmacytic infiltrate, suggestive of chronic endometritis. There was no cytological atypia [Figure 2]. Following the procedure, the patient was lost to follow-up.

DISCUSSION

Endometrial osteocartilaginous metaplasia is a benign entity characterized by the presence of bone and cartilage within the endometrium. Rare cases of endometrial osseous metaplasia and endometrial cartilaginous metaplasia have previously been reported;[3-7] however, no previously published case reports of combined endometrial osteocartilaginous metaplasia were found on searching the literature.

The exact etiology of endometrial ossification is unknown; however, there are several theories which have been proposed. These include the retention of fetal bones following abortion with secondary promotion of osteogenesis in the surrounding endometrium;[8] dystrophic calcification of retained and necrotic tissues, usually after an abortion; osseous metaplasia of endometrial stroma in response to chronic endometrial inflammation and the reparative process induced by abortion;[9] osseous metaplasia from multipotential stromal cells which become osteoblasts;[10] continuous and strong endometrial estrogenic stimulation; and metabolic disorders such as hypercalcemia, hypervitaminosis D, or hyperphosphatemia.[4]

Reported cases of osseous metaplasia frequently have a history of previous pregnancy loss and hence the proposal that this entity is most likely related to embedded fetal tissue following pregnancy termination. However, DNA analysis of a recent case confirmed that osseous metaplasia was true metaplasia and not related to fetal remnants.[11] Patients typically present with secondary infertility, menstrual irregularities, pain, or dysmenorrhea.[9,12] Ultrasound examination shows a characteristic hyperechogenic pattern which is strongly suggestive of endometrial ossification.[4] This may be confirmed by hysteroscopic examination and subsequent histological examination of the lesion. Histological examination shows endometrial tissue containing foci of bone which blend with the endometrial cells, indicating origin from the stromal cells. Cartilaginous metaplasia is even rarer but shows similar histological features with islands of cartilage, which merges with endometrial stromal cells at the periphery. This is in contrast to retained cartilaginous material from an aborted fetus in which transition from stromal cells is not seen.

As islands of bone and cartilage can represent heterologous elements of malignant mixed Mullerian tumor, careful histological assessment of the surrounding stromal cells for the presence of cytological atypia is important. The failure to recognize the existence of benign endometrial osteocartilaginous metaplasia will result in overdiagnosis and inappropriate treatment.

Other forms of mesenchymal metaplasia seen within the endometrium include myomatous metaplasia, adipose metaplasia, and synovial metaplasia. Stromal myomatous nodules compressing surrounding glands have been seen. This is commonly associated with high-potency

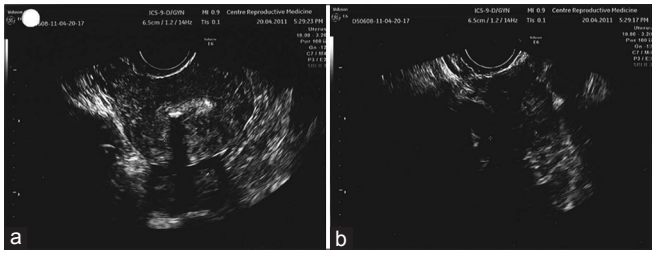

Figure 1: Transvaginal ultrasound with 14 MHz probe demonstrating a calcified endometrial polyp, (a) (see acoustic shadowing b)

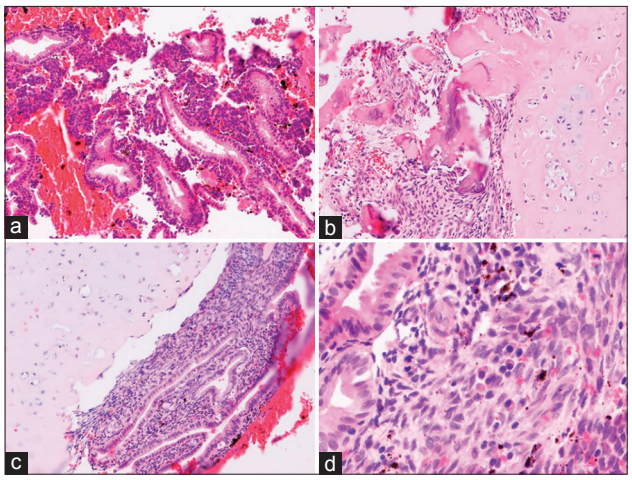

Figure 2: (a) Blood admixed with fragmented endometrial tissue showing stromal clumping and glandular disintegration (H and E, ×20). (b) Mature bone and cartilage (H and E, ×20). (c) Endometrium and an island of cartilage (H and E, ×20). (d) Plasmacytic infiltrate within the endometrium (H and E, ×60)

progesterone therapy.[13] These have been regarded as endometrial leiomyomas but are probably metaplastic in origin.[14] Adipose metaplasia rarely occurs in nonpolypoidal endometrium, and the recognition of this entity in a curettage specimen will avoid misdiagnosing uterine perforation and the inclusion of extrauterine adipose tissue, causing unnecessary anxiety to both the patients and clinicians.[15] Synovial-like metaplasia has been described by Stewart *et al*. This is a focal finding associated with surface epithelial erosions seen in endometrium associated with the levonorgestrel-releasing intrauterine system. A variable proportion of the cells express CD68 and are regarded as modified fibroblasts or stromal cells.[16]

Since osteocartilaginous metaplasia may have a similar appearance to mature heterologous elements of a malignant mixed Mullerian tumor, it is important for histopathologists to be aware of this entity to avoid misdiagnosis. The gold standard for the treatment of osseous metaplasia is hysteroscopic removal of the lesion.[6]

CONCLUSION

It is important for histopathologists to be aware of these rare and benign conditions, in particular, endometrial osteocartilaginous metaplasia, to avoid the misdiagnosis of a malignant mixed Mullerian tumor, with consequent inappropriate investigation and treatment. Hence, all endometrial biopsies containing areas of metaplasia must, therefore, be interpreted with great caution and in the context of appropriate clinical and demographic information to exclude more ominous pathology.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names 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.

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