

A Rare Case Of Endometrial Ossification

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Citation

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Abstract

We report a rare case of endometrial ossification in a 28 year old female, who presented with abdominal pain, menorrhagia and secondary infertility. On transvaginal ultrasonography, a hyper-echoic area within the uterine cavity, suggestive of an intra-uterine foreign body was noted. Histopathological examination of the endometrial curettage showed bony spicules with inflammatory cells infiltration. There was no evidence of malignancy.

INTRODUCTION

Endometrial osseous metaplasia, an uncommon entity which is related to secondary infertility following an abortion, is the presence of mature and immature bone in the endometrium. Less than 100 cases have been reported in the world literature, including nine from India [1],[2],[3]. Most of the patients conceive after the hysteroscopic evacuation of the bony spicules [4],[5]. We are reporting a case of endometrial ossification in a 28 year old female, who presented with menorrhagia and secondary infertility.

CASE SUMMARY

A 28 year old female patient presented to Gynecological Out Patient Department with the complaints of abdominal pain, menorrhagia and secondary infertility. Her past history revealed that she had one living child and two repeated abortions 6 years back, after which she had failed to conceive. Dilatation and curettage was done at 4 months of gestation during the last conception. The patient had no known history of systemic disease. The patient had no signs or laboratory findings which suggested a calcium metabolism disorder. Her serum calcium and phosphorus levels were normal. Further evaluation by ultrasonography revealed a densely echogenic band occupying most of the endometrial cavity. The patient underwent diagnostic hysteroscopy which revealed a single firm to hard tissue piece within the endometrial cavity, which were removed by using hysteroscopic forceps and these were submitted for histopathological study.

Grossly, the biopsy specimen included multiple, small, firm to hard tissue bits, along with scanty soft tissue pieces, together measuring 1×0.5×0.5 cms in dimension.(figure 1)

The hard tissue bits were kept for decalcification. The hematoxylin and eosin stained paraffin sections were subjected for microscopic examination. The sections showed fragmented endometrial tissue which was predominantly composed of tubular glands with scanty stroma. The osteoid tissue which was mainly composed of the trabeculae of woven bone was present surrounding the endometrial tissue and inflammatory cells. The endometrial glands did not show any secretory activity. Further examination did not reveal any granuloma, necrosis or the products of conception. The histological diagnosis of osseous metaplasia of the endometrium was made.(figure 2)

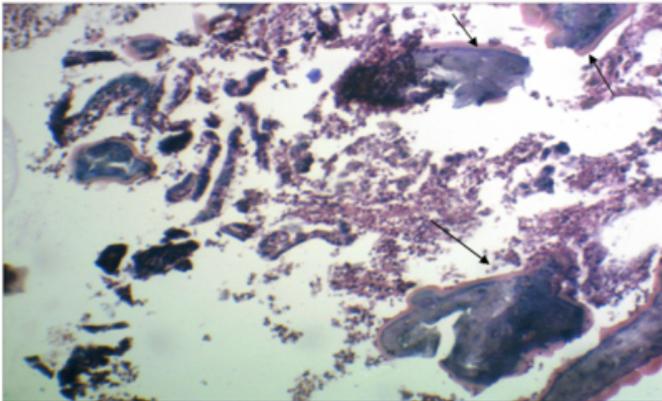
Figure 1

Figure 1: Gross appearance of the endometrial curettage



Figure 2

Figure 2: Microscopic appearance (H & E stain x 100) arrows pointing ossified spicules



DISCUSSION

Endometrial ossification is an uncommon disease related to secondary infertility and its etiology and pathogenesis are controversial. More than 80% of reported cases occur after pregnancy [6]. The most widely accepted hypothesis is that ossification represents retained fetal bones following spontaneous, missed, incomplete or therapeutic abortion, suggesting endochondral ossification. It can also be related to transformation of mesenchymal tissue to bone in response to inflammation and the reparative process induced by abortion [7]-[9].

Many theories have been proposed: osseous metaplasia from multipotential stromal cells, usually fibroblasts, which become osteoblasts [10]; continuous and strong endometrial estrogenic stimulation; retention of fetal bones that secondarily promote osteogenesis in the surrounding endometrium [11]; implantation of embryonic parts without pre-existing bone after abortions at an early stage; dystrophic calcification of retained and necrotic tissues, usually after an abortion; chronic endometrial inflammation such as endometritis or pyometra and metabolic disorders such as hypercalcemia, hypervitaminosis D or hyperphosphatemia. [12] The actual contribution of these pathogenic mechanisms is unknown [13]. In our case, there was no evidence of a calcium metabolic disorder. Bhatia and Hoshiko reported a case of osseous metaplasia involving both the endometrium and the endocervix. They believed this could be associated with prolonged chronic inflammation and tissue destruction following repeated spontaneous or therapeutic abortions. Foetal bones may serve as a source of calcium for ossification, but this may be valid only for abortions occurring in the second trimester, when ossification of the

foetal skeleton has reached a certain level. [14]. According to Marcus et al, the reactive endometritis probably caused by the presence of the bone fragments interferes with blastocyst implantation. [12] Ectopic bone formation and calcification result from the insult of chronic inflammation or tissue destruction with repeated abortions [15]. Also supporting the presence of inflammation in cases of endometrial osseous metaplasia, that the removal of bone fragments from the endometrium in these cases reduced the local concentrations of prostaglandin in 50%, as it has been documented by Lewis et al. [16] In our case, the endometrial curettage showed evidence of chronic nonspecific inflammation.

A previous history of abortion is present in most of the reported cases with osseous changes in the endometrium, as was seen in our case. Usually the reproductive age group is involved, with a history of first trimester abortion. These patients resume the normal menstrual cycle in the post abortive period, as was seen in our case. The time interval between the antecedent abortion and the discovery of the endometrial ossification varies from 8 weeks to 14 years in the reproductive age group. In our case, the time interval was 6 years from the abortion, as compared to the history of 37 years from the abortion, which was described by Shimazu and Nakayama in a 62 years old woman [4],[17]

Physicians should also be aware of osseous metaplasia in the differential diagnosis of patients with uncertain history, who present with a sonographic image resembling an intrauterine contraceptive device [3],[4]. In our case, there was no history of use of intrauterine contraceptive device.

Before classifying the heterologous tissue as benign, the pathologist should exclude the possibility that the tissue in question is not a deceptively bland appearing component of a malignant mixed Mullerian tumor or an adenosarcoma. In our case no such tumor elements were seen.

Endometrial tuberculosis should be ruled out, as it is a common cause of infertility in Indian females and can sometimes cause calcification and subsequent ossification in the endometrium. In our case, as there was no evidence of granuloma or caseous necrosis, tuberculosis was ruled out.

Retained foetal tissue is also an important cause for osseous metaplasia. The absence of surrounding tissue reaction and endochondral ossification may differentiate osseous metaplasia from the retained foetal tissue [4]. In our case, as there was history of abortion, 6 years back, retained foetal tissue during this conception could be the reason for

endometrial ossification.

The removal of the heterotopic tissue is expected to restore the fertility with good endometrial response to the cyclical stimulation.[18]

CONCLUSION

This case report highlights endometrial ossification as a very rare and peculiar cause of infertility. This might be due to retained products of conception during the last abortion, which took place 6 years back. Fertility can be restored by the complete removal of the bony spicules from the endometrial cavity. Malignant mixed Mullerian tumor and endometrial tuberculosis are other important causes of ossification in endometrial cavity. Osseous metaplasia in endometrial cavity can resemble Intrauterine contraceptive device on sonography. So, clinicians and pathologists should be aware of this entity in order to avoid an erroneous diagnosis.

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