

Endometrial ossification: A rare occurrence with review of literature

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ABSTRACT

Endometrial ossification is a rare occurrence seen in reproductive age group. Patients usually present with complaints of menorrhagia and secondary infertility. We report a case of endometrial ossification in a 28-year-old female who presented with abdominal pain and menorrhagia. On transvaginal ultrasonography, a hyperechoic area within the uterine cavity, suggestive of an intrauterine foreign body was noted. Histopathological examination of the endometrial curettage showed bony spicules with chronic inflammatory cell infiltration.

Key words: *Bony, Endometrial, Infertility, Menorrhagia, Ossification, Spicules*

Endometrial ossification is an uncommon entity which is related to secondary infertility following an abortion. Its etiopathogenesis is controversial, and more than 80% of the reported cases occur after pregnancy [1]. The most widely accepted hypothesis is that ossification represents retained fetal bones following spontaneous, missed, and 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 <100 cases have been reported in the world literature [2]. The gold standard for treatment is hysteroscopy. Most of the patients conceive after hysteroscopy evaluation of bony spicules. We present a rare case report with review of literature on endometrial ossification in a 28-year-old female patient who presented with pain abdomen and menorrhagia.

CASE REPORT

A 28-year-old female patient presented to the gynecological outpatient department with complaints of abdominal pain and menorrhagia. She had one living child and had undergone medical termination of pregnancy of 2 months duration. She had no history of tuberculosis or any systemic medical illness in the past. The patient also had no signs or laboratory findings which suggested a calcium metabolic disorder. Her serum calcium and phosphorous levels were normal. Further evaluation by ultrasonography revealed a densely echogenic band filling the endometrial cavity. Dilatation and curettage were done.

Grossly, the biopsy specimen included multiple, small, firm to hard tissue bits along with scanty soft tissue pieces, together measuring 1 cm × 0.5 cm × 0.5 cm. The hard tissue bits altogether measured 0.5 cm × 0.4 cm × 0.2 cm and were kept for

decalcification. The hematoxylin and eosin (H and E) sections were subjected to microscopic examination. The sections showed fragmented endometrial tissue with dense inflammatory infiltrate comprising of plasma cells and lymphocytes. A focus of osteoid matrix was seen. No decidua, villi, or trophoblastic tissue was found on serial sectioning. Sections from the bony bit show V of woven bone enclosing marrow. The endometrial glands did not show any secretory activity. Further examination failed to reveal any granuloma, necrosis, or products of conception (Figs. 1-5). The histological diagnosis of osseous metaplasia of endometrium was made. After dilatation and curettage, patient was relieved of pain abdomen and menorrhagia and is doing fine on follow-up.

DISCUSSION

Endometrial ossification is an uncommon entity. More than 80% of reported cases occur after pregnancy [1]. Patients present with complaints of infertility, heavy menstrual bleeding, and pain abdomen. Its etiology and pathogenesis are controversial [2]. The most widely accepted hypothesis is that ossification represents retained fetal bones following spontaneous, missed, incomplete or therapeutic abortion, and suggesting endochondral ossification. Another hypothesis is the transformation of mesenchymal tissue to bone in response to inflammation and the reparative process induced by abortion [3]. Many theories such as osseous metaplasia from multipotent stromal cells have been proposed such as fibroblasts which become osteoblasts, continuous, and strong endometrial estrogenic stimulation, retention of fetal bones that secondarily promote osteogenesis in endometrium, dystrophic calcification of the retained and necrotic tissues after abortion, chronic endometrial inflammation such as endometritis

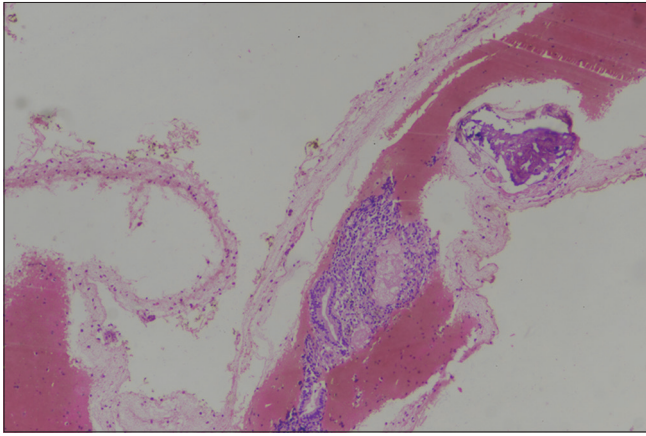


Figure 1: Photomicrograph showing endometrial gland with woven bone formation in hemorrhagic background (H and E; ×40)

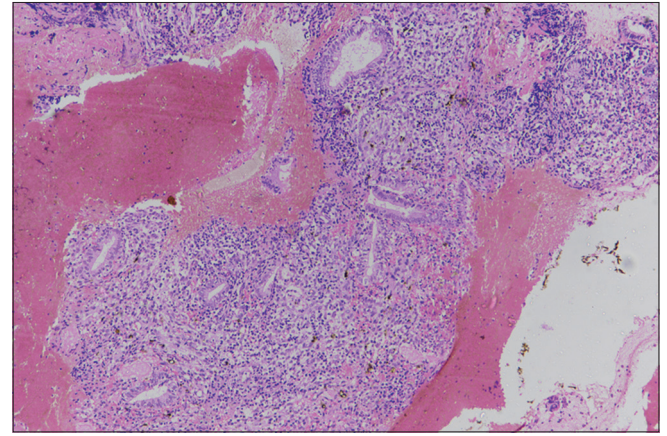


Figure 4: Photomicrograph showing endometrial glands and stroma packed with dense chronic inflammatory cells and hemorrhage (H and E; ×100)

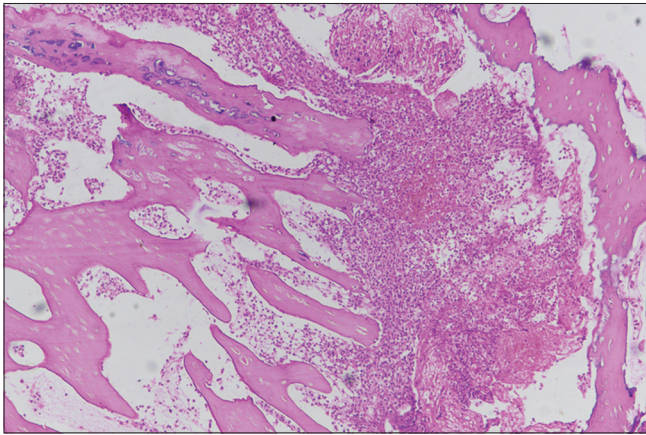


Figure 2: Photomicrograph showing bony trabecular with hematopoietic cells (H and E; ×100)

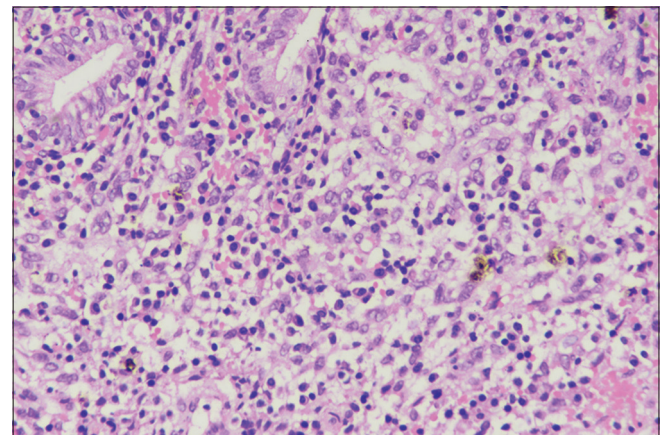


Figure 5: Photomicrograph showing endometrial glands and stroma packed with dense chronic inflammatory cells in higher magnification (H and E; ×400)

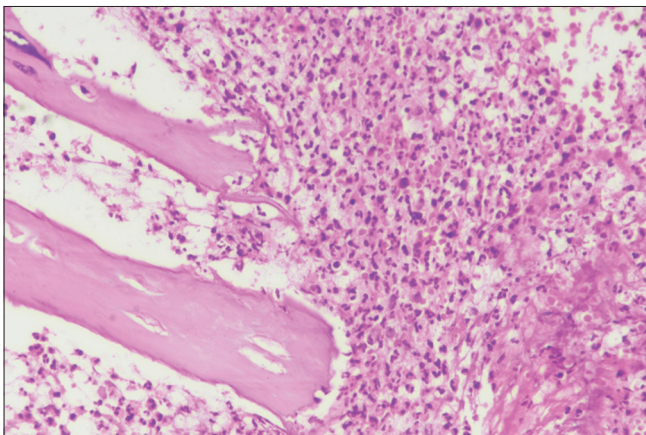


Figure 3: Photomicrograph showing bony trabecular with hematopoietic cells in higher magnification (H and E; ×400)

or polymeric and metabolic disorders such as hyperkalemia, hypervitaminosis D, or hypophosphatemia [4-7]. In our case, there was no clinical or biochemical evidence of calcium metabolic disorder. Bhatia and Hoshiko reported a case of osseous metaplasia involving both endometrium and cervix [8]. They believed this could be associated with chronic inflammation and tissue destruction following repeated spontaneous or therapeutic abortions. Fetal bones serve as a source of calcium for ossification

if abortions occur in the second trimester, when ossification of the fetal skeleton has reached [9]. According to Marcus et al., the reactive endometritis probably caused by the presence of the bone fragments interferes with blastocyst implantation [10]. Ectopic bone formation and calcification result from the insult of chronic inflammation or tissue destruction with repeated abortions.

A prior history of abortion is present in most of the reported cases with osseous changes in the endometrium as is seen in our case. Usually, the reproductive age group is involved with a history of first-trimester abortion. [10,11]. The time interval between the antecedent abortion and the discovery of the endometrial ossification varies from 8 weeks to 14 years. In our case, the time interval was 2 months from the time of abortion as compared to the history of 6 and 37 years from abortion which was described by Virchow and Shimazu and Nakayama, respectively [12-14]. A comparative study of various previously reported cases of endometrial ossification is given in Table 1. Clinicians should 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. In our case, there was no history of use of intrauterine contraceptive device [15]. Before classifying the heterologous tissue as benign,

Table 1: A comparative study of various reported cases of endometrial ossification

Case description	Number of cases	Name of study	Year of study
Patient presented with infertility	01	Rodriguez and Adamson [9]	1993
Patients presented with metro menorrhagia and secondary infertility	02	Marcus et al. [10]	1994
Patient with secondary infertility after an abortion 12 years back	01	Bahçeci and Demirel [4]	1996
Patient with metrorrhagia and leucorrhoea	01	Ceccacci and Clancy [7]	1996
Patient with dysmenorrhoea with previous 2 abortions	01	Torné et al. [13]	1996
Patient with diagnosis of endometrial osseous metaplasia 20 days after spontaneous delivery	01	Van den Bosch et al. [17]	2000
Case report of endometrial osseous metaplasia associated with secondary infertility	01	Lainas et al. [6]	2004
Patient with secondary infertility after two abortions	01	Onderoglu et al. [18]	2008
28-year-old lady with menorrhagia and history of previous abortion 2 months back	01	Present study	2017

the pathologists should exclude the possibility that the tissue in question is not deceptively bland appearing component of a malignant mixed Müllerian tumor or adenosarcoma [16]. In our case, there was no such tumor elements noted.

Endometrial tuberculosis should also be ruled out as it is a common cause of infertility in developing countries like India and can sometimes lead to calcification and subsequent, ossification in the endometrium [17]. In our case, there was no evidence of epithelioid cell granuloma or caseous necrosis; hence, tuberculosis was excluded from the study. Retained fetal tissue is also an important cause for osseous metaplasia. The absence of surrounding tissue reaction and enchondral ossification may differentiate osseous metaplasia from retained fetal tissue [18]. In our case, there was a history of abortion 2 months back, retained fetal tissue during this conception could be the reason for endometrial ossification; however, no chorionic villi or trophoblastic tissues were seen on histopathology.

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 past abortion, which took place 2 months back. Fertility can be restored by the complete removal of the bony spicules from the endometrial cavity. Osseous metaplasia in endometrial cavity can resemble intrauterine contraceptive device on ultrasonography. Hence, clinicians and pathologists should be aware of this entity to avoid an erroneous diagnosis.

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