

CASE REPORT

Osseous Metaplasia of Endometrium: A Very Rare Entity

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ABSTRACT

Introduction: Osseous metaplasia of endometrium is a rare disorder characterized by the presence of mature or immature bone in the endometrium. The rarity of the case and high probability of missing out, which may lead to delay in appropriate treatment, makes this case of interest.

Case report: A 37-year-old lady, married for 10 years P2L2 with previous two lower-segment cesarean section, presented to the gynecology outpatient department with complaints of secondary infertility for past 7 years, irregular menstrual cycles, dysmenorrhea, and dyspareunia. Her general examination and bimanual exploration were normal. On ultrasonography, an echogenic foci was seen casting posterior acoustic shadow. On diagnostic laparohysteroscopy, multiple small coral-like white plaques—bony spicules were seen. They were removed using hysteroscopic forceps and submitted for histopathological study. A histological diagnosis of osseous metaplasia of endometrium was made. She is on follow-up for infertility treatment.

Conclusion: Endometrial ossification is a rare finding, which can be misdiagnosed and requires higher degree of suspicion to diagnose the condition properly. Hysteroscopy has been shown to be effective in the diagnosis and treatment of cases of osseous metaplasia of the endometrium associated with infertility.

Keywords: Hysteroscopy, Infertility, Osseous metaplasia.

How to cite this article: Gupta G, Mahindru D, D'Souza A, Goyal S. Osseous Metaplasia of Endometrium: A Very Rare Entity. *World J Lap Surg* 2017;10(2):69-72.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Osseous metaplasia of endometrium is a rare¹ disorder characterized by the presence of mature or immature bone in the endometrium. It is a rarely encountered condition, with an estimated incidence of 3/10,000.² Nearly 80 cases have been reported in the world literature, including around 9 cases from India.³

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It appears in women of reproductive age, though it has been reported in menopausal women as well. A history of previous pregnancy is reported in more than 80% of the cases.⁴ A history of abortion, either spontaneous or therapeutic, is the hallmark of this condition.⁵ It can also be related to transformation of mesenchymal tissue to bone in response to inflammation and the reparative process induced by abortion.

Most cases present with secondary infertility following an abortion or chronic endometritis. Some patients are asymptomatic, while others have menstrual irregularities or menorrhagia. Ultrasound examination showing characteristic hyperechogenic pattern of osseous tissue within the uterus helps suspect the diagnosis. The final diagnosis is confirmed by hysteroscopy and removal of the bony tissue by curettage. Complete removal of the bony spicules from the endometrial cavity by hysteroscopy usually cures the patient.

CASE REPORT

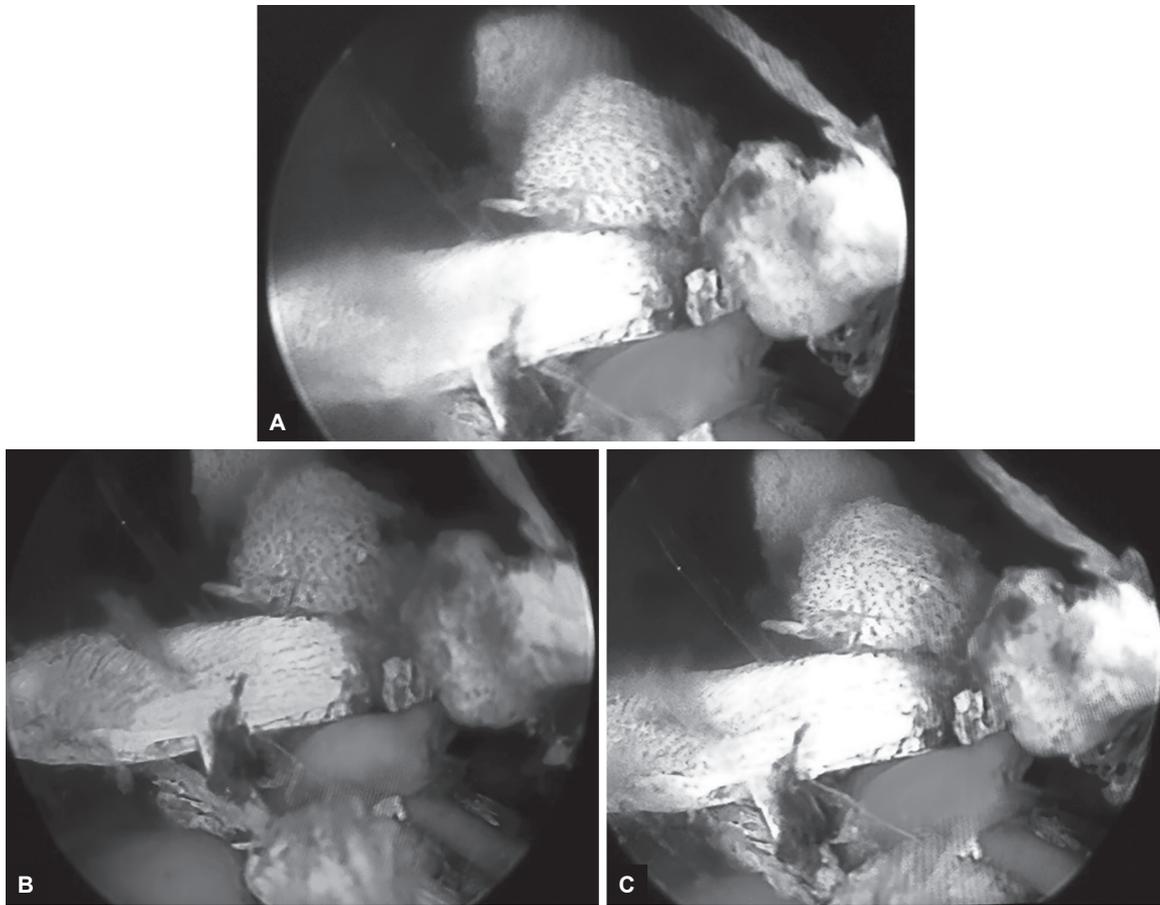
A 37-year-old lady married for 10 years P2L2 with previous two lower-segment cesarean section presented to the obstetrics and gynecology outpatient with complaints of secondary infertility for past 7 years, irregular menstrual cycles with intermenstrual bleeding. She presented with history of dysmenorrhea and dyspareunia.

Her general examination and bimanual exploration were normal. Her routine hematological parameters were normal. Both couples were subjected for infertility workup and nothing abnormal was noted.

Ultrasound pelvis showed the uterus to be of 5.2 × 4.3 cm size with normal shape, and echotexture. Endometrial stripe was 7.5 mm. An echogenic foci was seen casting posterior acoustic shadow. Cervix was normal with closed internal os. Bilateral ovaries were normal. No adnexal mass or free fluid was noted in pouch of douglas.

She was taken up for diagnostic laparoscopy and chromohysteroscopy. On laparoscopy, uterus was found to be bulky with normal bilateral tubes and ovaries. Chromopertubation test showed bilateral free spill, suggestive of patency of both fallopian tubes.

Hysteroscopy was done using rigid hysteroscope with saline as distending media and multiple small coral-like white plaques – bony spicules were seen. They were removed using hysteroscopic forceps and submitted for histopathological study (Fig. 1).



Figs 1A to C: Hysteroscopic appearance of endometrial osseous metaplasia showing multiple coral like white plaques

Histopathological examination revealed the following:

- Stroma with interspersed fragmented endometrial glands (secretory phase)
- Cells with vacuolations along with spicules of lamellar bone
- No inflammatory cell infiltrates
- No granulomas
- No chorionic villi or fetal remnants

A histological diagnosis of osseous metaplasia of endometrium was made. She made an uneventful recovery (Fig. 2).

DISCUSSION

Osseous metaplasia of the endometrium is a rare clinical entity characterized by the presence of mature or immature bone in the endometrium. It is described as an endogenous nonneoplastic pathological condition as no tissue reaction is found in the endometrial tissue studied and the endometrium showed normal regular cyclical changes. Its estimated incidence is 3/10,000,² there being about 80 cases described in the literature.³

It has been referred to by various names like ectopic intrauterine bone, heterotopic intrauterine bone, endometrial ossification, etc.¹

Majority of the patients belong to the reproductive age group with history of first trimester abortion, either

therapeutic or spontaneous, and have normal menstrual cycle in the postabortive period. The interval between the antecedent pregnancy and detection of endometrial ossification varies from 8 weeks to 14 years.⁶

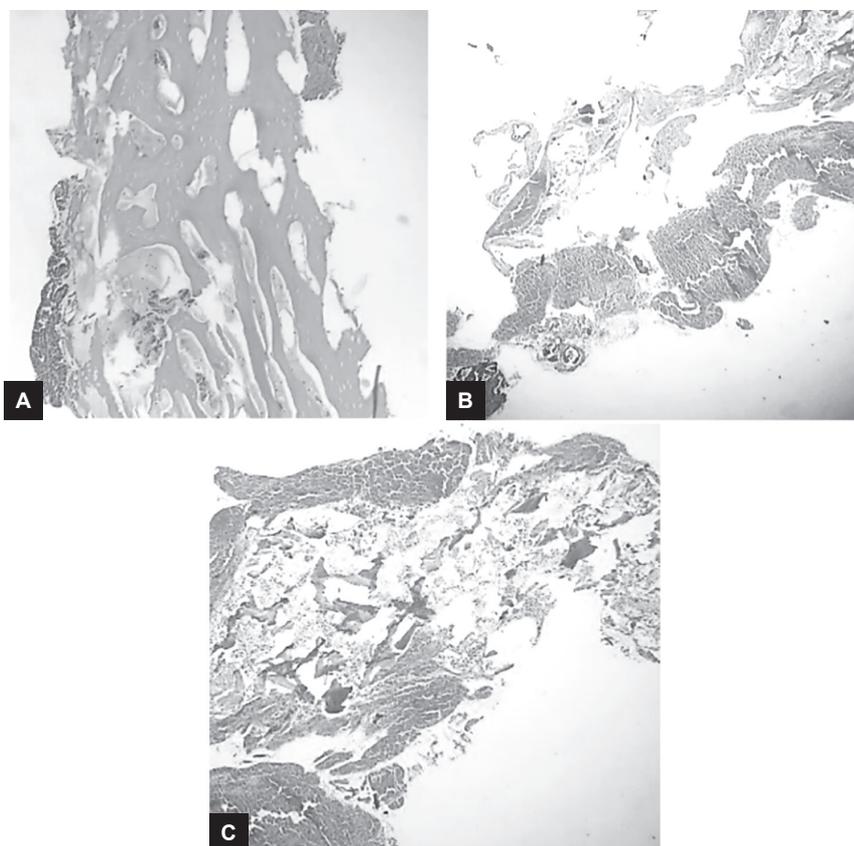
Pathogenic mechanisms related to the histogenesis of heterotopic bone into the endometrium are controversial. As early as 1884, Virchow attributed the formation of bone in the endometrium to spontaneous differentiation of fibroblasts into osteoblasts.⁷ Various theories have been described in the pathogenesis of endometrial osseous metaplasia.

The most common theories proposed are heterotopia, dystrophic calcifications, ossification of postabortive endometritis, metastatic calcification, metaplasia in healing tissue, prolonged estrogenic therapy after abortion, and retained fetal bone.

Genital tuberculosis, unspecific chronic endometritis, or pyometra are other sources of chronic inflammation occurring from retained embryonic tissue after first trimester abortion. This inflammation acts as a promoter of secondary osteogenesis from nonosseous embryonic tissue.⁸

In India, endometrial tuberculosis should be ruled out as it can cause infertility as well as calcification and subsequent ossification.¹

Chronic endometritis stimulates the proliferation of mesenchymal cells that have inherent property of



Figs 2A to C: Histopathological appearance of endometrial osseous metaplasia showing secretory phase stroma with interspersed endometrial glands and vacuolated cells with spicules of lamellar bone

metaplasia and can differentiate into chondroblasts or osteoblasts.¹

Adamson linked endometrial ossification to hyper-*vitaminosis* in one of his two patients reported to have ossification after therapeutic abortions.²

It is also probable that the concept of a superoxide radical superoxide dismutase system, which plays an important role in endometrial differentiation, may be functional in osseous metaplasia. Chronic postabortal inflammation due to retained gestational tissues may promote superoxide radical or tumor necrosis factor release from thernuclear phagocytes. Endometrium deficient in protective superoxide dismutase activity may perhaps present a long-lasting insult to the multipotential stromal cells, and this insult may therefore transform these cells into osteoblasts.⁹

Roth and Taylor¹⁰ demonstrated the presence of acid mucopolysaccharides, thereby supporting the capability of mature endometrial stromal cells to undergo cartilaginous metaplasia in response to chronic inflammation or trauma.

Use of estrogen is controversial as it can promote osteogenesis and can be one of the causes of endometrial ossification.

The most common presentation of osseous metaplasia of endometrium is usually secondary infertility. The

patients can also present with menstrual irregularities, pelvic pain, dyspareunia, and vaginal discharge. Osseous metaplasia causes subfertility and menstrual irregularities by changing the *milieu* of uterine cavity through the increased production of prostaglandins. Marcus et al¹¹ proposed increased prostaglandin production as a cause of subfertility in the presence of bony fragments.

Ultrasound examination plays a primary role in the diagnosis of patients with osseous metaplasia.⁷ The characteristic hyperechogenic pattern is strongly suggestive of osseous tissue within the uterus and should be confirmed by hysteroscopic examination. The gold standard for its diagnosis and treatment is hysteroscopy.¹² Hysteroscopy is an effective means of extracting the heterotopic tissue from the uterus and reestablishing fertility, even after a long period of infertility.

Complete removal of the bony spicules from the endometrial cavity by hysteroscopy regains fertility and cures menstrual symptoms.

CONCLUSION

Endometrial ossification is a rare entity, which can be misdiagnosed and requires higher degree of suspicion to diagnose the condition properly. Sonography plays an important role in detecting this condition. Osseous metaplasia can be deeply embedded in the uterine

mucosa and may present the same contraceptive effect as an intrauterine contraceptive device, thereby causing secondary infertility. Hysteroscopy has been shown to be effective in the diagnosis and treatment of cases of osseous metaplasia of the endometrium associated with infertility.

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