



Endometrial Osseous metaplasia associated with secondary infertility - Report of 2 cases

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Abstract : Endometrial osseous metaplasia is a rare clinical entity with the presence of mature or immature bone in the endometrium. Nearly eighty cases have been reported in the world literature. It can cause secondary infertility and it occurs in more than eighty percent of patients after an abortion. Here is a report of two cases of secondary infertility who came for evaluation and found to have osseous metaplasia on workup. Case 1- A thirty four years old nulliparous lady, married for six years with history of a spontaneous abortion at eight weeks of gestation. Ultrasound showed small subendometrial and endocervical calcific foci. . Dilatation and curettage was done and endometrial curettings were sent for histopathological examination which revealed foci of heterotopic ossification. Case 2 - A thirty years old nulliparous lady presented with secondary infertility. She gave history of spontaneous abortion at ten weeks of gestation, 4 years back. Ultrasound pelvis revealed a linear hyperechoic area of 8mm length in lower endometrial cavity. Hysteroscopic examination was done and endometrial curettings were taken. The histopathology report showed osseous metaplasia. These case reports highlight peculiar cause of infertility and discusses its management by hysteroscopy. Osseous metaplasia of endometrium is described as an endogenous non-neoplastic pathological condition. Its estimated incidence is 3 out of 10000 people. Heterotopia, dystrophic calcification, ossification of post-abortion endometritis, metastatic calcification, metaplasia in healing tissue, prolonged estrogenic therapy after abortion and retained fetal bone are the commonly proposed theories. In most of the reported cases, the osseous changes followed a previous history of abortion. The time interval between the antecedent abortion and discovery of the ossification varies from 8 weeks to 14 years. Ultrasound examination plays a primary role in the diagnosis of patients with osseous metaplasia. However, it should be confirmed by hysteroscopic examination.

Keyword : Endometrial metaplasia, secondary infertility, abortion, osteoblastic cells.

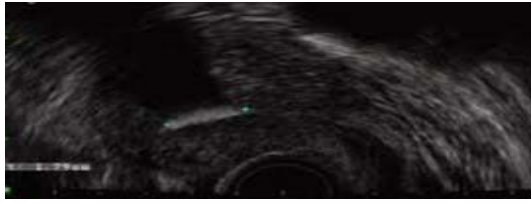
INTRODUCTION:

Endometrial osseous metaplasia is a rare clinical entity characterized by the presence of mature or immature bone in the endometrium. Nearly eighty cases have been reported in the world literature. Most patients are asymptomatic while a few others present with menstrual irregularities or menorrhagia. It occurs in more than eighty percent of patients after an abortion and hence it can cause secondary infertility. Various theories have been proposed and the most accepted one is "metaplasia of the stromal cells into osteoblastic cells that produce bone". Ultrasound examination shows characteristic hyper-echogenic pattern of osseous tissue within the uterus and this helps to suspect the diagnosis. The final diagnosis is confirmed by hysteroscopic removal of the bony tissue and its histopathological report. The following is a case report of two patients who presented with secondary infertility.

CASE REPORT:

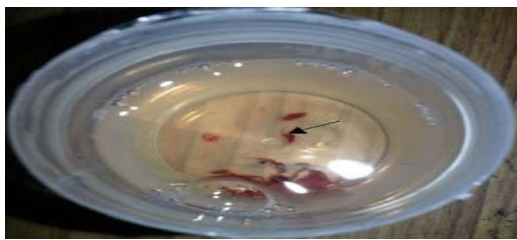
CASE 1:

A 34 years old lady who is married for six years presented to gynecology out-patient department with secondary infertility. She gave history of a spontaneous abortion at eight weeks of gestation, two years back. She was anxious to conceive and hence had come for investigating secondary infertility. Her bimanual examination was normal. Uterus was normal in size, fornices were free and there was no fullness or tenderness. Her routine investigations like complete hemogram, blood sugar, TSH and prolactin were normal. Her husband's semen analysis was normal. Her pelvic ultrasound showed uterus measuring 9.2 x 6.3 x 4.2 cm with small subendometrial and endocervical **calcific foci**; both ovaries showed polycystic features. Diagnostic laparoscopy was done on day 5 of cycle. Bilateral tubal spill was present. Both ovaries were polycystic. Dilatation and curettage was done under ultrasound guidance, expecting some difficulty. But there was no difficulty. Moderate quantity of endometrial curettings were obtained and were sent for histopathological examination. Histopathological report revealed: **Secretory endometrium with foci of heterotopic ossification.** The patient did not turn up for follow up and could not contact her.

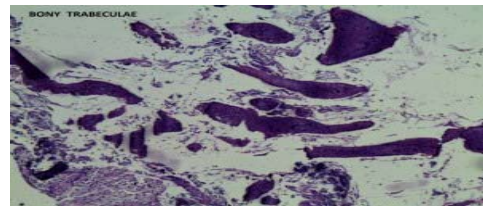


CASE 2:

A 30 years old lady, presented to our gynecology outpatient department with secondary infertility. She is married for 4 years. Her menstrual cycles were regular, normal flow with dysmenorrhea on first day. She had conceived soon after marriage but it ended in a spontaneous abortion at 10 weeks of gestation, which was completed by dilatation and curettage. After that she underwent 2 to 4 cycles of ovulation induction with clomiphene citrate with documented follicular rupture but failed to conceive during the treatment. Her husband's semen analysis was normal. Her general and systemic examinations were normal. Her body mass index was 29. Bimanual examination was done. Uterus was of normal size, retroverted and the fornices were free. Investigations including complete hemogram, blood sugar, TSH, Prolactin, FSH and LH were within normal limits. Her pelvic ultrasound revealed normal sized uterus with a linear hyper echoic area of 8mm length in lower endometrial cavity. Endometrial thickness was 6mm. Bilateral ovaries were polycystic. Hysteroscopy was done. In hysteroscopic view a thin whitish tissue was visualized over posterior endometrium; the same was removed and sent for histopathological examination. Through laparoscopy, tubal patency was checked by chromotubation and there was tubal spill on both sides. Both ovaries were polycystic.



Histopathological report of the endometrial tissue showed: **Non Proliferative endometrium with osseous metaplasia**. She is on follow up and not yet conceived. She has been advised on life style modification with weight reduction.



DISCUSSION:

Osseous metaplasia of endometrium is described as an endogenous non-neoplastic pathological condition. Its estimated incidence is 3/10000. WHO classification of endometrial metaplasia and related changes[1]

1. Epithelial metaplasia
2. Non-epithelial metaplasia
 - Smooth muscle metaplasia
 - **Osseous metaplasia**
 - Cartilaginous metaplasia
 - Fatty change
 - Glial tissue
 - Foam cell change

Heterotopia, dystrophic calcifications, ossification of post-abortive endometritis, metastatic, calcification, metaplasia in healing tissue, prolonged estrogenic therapy after abortion and retained fetal bone are the commonly proposed theories. [2][3] Bahceci and Demirel suggested that post abortive chronic endometritis stimulates the release of superoxide radicals and tumor necrosis factor from the inflammatory cells. Their long term exposure on multipotent stromal cells in patients with deficient superoxide dismutase activity in the endometrium leads to metaplasia of the stromal cells into osteoblastic cells. [4] In most of the reported cases, the osseous changes followed a previous history of abortion. [5][6] The time interval between the antecedent abortion and discovery of endometrial ossification varies from 8 weeks to 14 years. [8] Common clinical presentations are menstrual irregularities, pelvic pain, dyspareunia, vaginal discharge and secondary infertility. [7] In both of our cases, secondary infertility was the presenting complaint. Ultrasound examination plays a primary role in the diagnosis.

The characteristic hyper echogenic pattern is strongly suggestive of osseous tissue within the uterus and should be confirmed by the hysteroscopic examination and histological evidence. Some suggest the use of estrogen for rapid endometrial regeneration but it is controversial as it is also known that estrogen may induce ossification. [7] Recent studies recommend hysteroscopic removal of the bony tissue or removal under ultrasonic guidance. [9] Bone in the endometrium can act as an intrauterine contraceptive device and its complete removal can restore the fertility and spontaneous conception [10]

CONCLUSION:

These case reports highlight a rare cause of infertility. Endometrial ossification is a rare but treatable cause of infertility in which intrauterine bone prevents normal conception. Review of literature shows that complete removal of the bony spicules from the endometrial cavity by hysteroscopy is the definitive treatment. Of these case reports, one of them lost follow-up. The other patient is still under treatment and follow-up.

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