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# **Case Report**

# The bony endometrium: a rare case of osseus metaplasia of endometrium

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## ABSTRACT

Osseous metaplasia of endometrium is an uncommon condition which presents as a bone in endometrium. The pathophysiology of this condition is still not understood completely. Many theories have been formulated to explain its occurrence. Most patients with this disorder present with 2° amenorrhoea or 2° infertility. Here, we present a case of 22 years old woman presenting to us with secondary amenorrhoea. Ultrasonography showed echogenic calcified endometrium suggestive of calcification. Hysteroscopy was done which revealed bony fragments in uterine cavity, which were removed, procedure went uneventful and patient was started on oestrogen and progesterone for endometrium regeneration. Histopathology report was suggestive of osseous metaplasia of endometrium.

Keywords: Osseous Metaplasia, Secondary amenorrhoea, Ultrasound, Infertility

#### **INTRODUCTION**

Endometrial osseous metaplasia is a rare entity characterised by the presence of mature or immature bone in the endometrium.<sup>1</sup> It is a rare condition, with <100 cases described in the literature, and an estimated incidence of  $3/10,000.^2$  Many theories have been put forward and the most accepted theory is metaplasia of the stromal cells into osteoblastic cells that produce mature bone.<sup>3</sup>

Here we present a case of osseous metaplasia of endometrium in a 22-year-old woman with secondary amenorrhoea managed hysteroscopically.

#### **CASE REPORT**

A -22-years-old female, with marital life of 5 years presented to our outpatient department with complaints of 3 years of amenorrhoea.

Patient had a full term normal vaginal delivery of a healthy child 4 years ago followed by a spontaneous abortion at 3 years back which was treated by dilatation and curettage. Her previous menstrual history was normal. There was no history of galactorrhoea, thyroid disorder, tuberculosis or any chronic medical/surgical illnesses. Her general, systemic and bimanual pelvic examination revealed no significant abnormality.

Husband semen analysis was within normal limits. Her routine haematological investigations (complete haemogram, fasting and post lunch blood sugar, liver and kidney function tests) and hormonal profile such as Thyroid stimulating hormone, prolactin, follicle stimulating hormone, and luteinizing hormone were within normal limits.

Ultrasonography of pelvis revealed a normal size uterus and adnexa with irregular areas of calcifications measuring  $18 \times 5.5$  mm with adhesions with no obvious infiltration into myometrium (Figure 1), features suggestive of calcified endometritis with differential diagnosis of endometrial tuberculosis.

Magnetic resonance imaging (MRI) abdomen and pelvis was ordered and results were consistent with ultrasound

report. Patient was posted for diagnostic laparoscopy and hysteroscopy. Tubal patency was confirmed by dye testing. Hysteroscopy was performed with saline as a distension medium, using a 4 mm Hopkins forward oblique 30° telescope (Karl Storz).

Hysteroscopy revealed multiple small bony fragments in the uterine cavity (Figure 2).

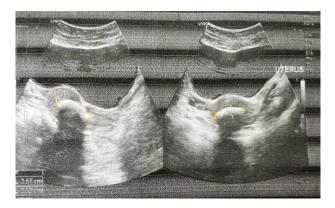


Figure 1: Ultrasonography of pelvis.



Figure 2: Multiple small bony fragments in uterine cavity in hysteroscopy.

Around 7-8 bony fragments of varying shapes and sizes were removed using a 5 Fr hysteroscopic grasper (Figure 3).



Figure 3: 7-8 bony fragments removed from the uterine cavity.

Uterine cavity was restored to normal. Post-operative period was uneventful without any complications.

Bony fragments were sent for histopathological examination, with suggested fragments of endometrial stroma mixed with fragments of mature bone tissue. Chronic villi or decidual tissue not identified suggestive of osseous metaplasia of endometrium.

Post procedure patient was started on cyclical estrogens and progesterone for 3 months and was asked to follow up for review hysteroscopy.

As patient failed to follow up, review hysteroscopy couldn't be done.

#### DISCUSSION

Osseous metaplasia of endometrium, also called as endometrial ossification, ectopic intrauterine bone, heterotopic intrauterine bone etc. is a rare clinical condition with <100 cases being reported in the literature.<sup>5</sup>

According to new World Health Organization (WHO) classification, endometrial metaplasia is divided into two categories i.e. with epithelial and non-epithelial changes.<sup>4</sup>

WHO classification of endometrial metaplasia and related changes- epithelial metaplasia and related changes, and non-epithelial metaplastic and related changes viz. smooth muscle metaplasia, osseous metaplasia, cartilaginous metaplasia, fatty change, glial tissue, and foam cell change.<sup>4</sup>

The clinical presentation of this symptom is very varied and patient may present with dysmenorrhoea, pelvic pain, menstrual irregularities, vaginal discharge, and secondary infertility or may remain asymptomatic.<sup>3,6</sup> Our case presented with complaint of secondary amenorrhoea for 3 years.

The probable mechanism for infertility or amenorrhoea is that the bony tissue in the uterus functions as intrauterine contraceptive device, thereby increase in the levels of prostaglandins and preventing blastocyst implantation.<sup>7</sup>

Most of the patients are in the reproductive age group with history of first trimester abortion either therapeutic or spontaneous.<sup>1</sup> The time interval from varies from 8 weeks to 14 years in reproductive age group, but it can be distinguished from retained fetal tissue by absence of tissue reaction and enchondral ossification.<sup>6,8</sup>

Several theories have been formulated that explain the presence of this tissue: metaplastic transformation of the endometrial stromal cell into osteoblastic cells that produce mature bone and implantation of fetal tissue after abortion and instrumentation with fetal tissue persisting and growing as a homograft.<sup>1,5</sup>

Estrogenic endometrial stimulation, chronic endometritis, pyometra leading to metaplasia metaplasia of uterine fibroblasts; dystrophic calcification in retained products of conception and post abortive endometritis are some other theories that have been made.<sup>1,3,6</sup>

According to study done by Sugino et al on changes in activity of superoxide dismutase in the human endometrium throughout the menstrual cycle and in early pregnancy, endometrium deficient in protective superoxide dismutase activity may perhaps present a long-lasting insult to the multipotential stromal cells, and this may therefore transform these cells into osteoblasts.<sup>11</sup>

In one case report done by Adamson and Sommers, endometrial ossification was found in a patient taking high dose of calcium and vitamin D for prolonged period, however association of osseous metaplasia of endometrium with hypercalcaemia and conditions causing hypercalcaemia is rare.<sup>2</sup>

Various genetic studies have also been done to determine the origin of endometrial bone; however, results have been different. According to study done by Cayuela et al and Parente et al, the bony tissue was of maternal origin, but in the study done by Tulandi et al the bony tissue was found to be of fetal origin.<sup>5,9</sup>

Endometrial tuberculosis, IUCD, malignant mixed mullerian tumour of endometrium and retained fetal bones after abortion are various differential diagnoses for this disorder.<sup>10</sup>

Hysteroscopy is considered the gold standard for definitive diagnosis and treatment of osseous metaplasia of endometrium. $^{1,6}$ 

In our case also hysteroscopic management was done and histopathology report was suggestive of osseous metaplasia of endometrium.

# CONCLUSION

Osseous metaplasia of endometrium is a rare pathological condition which is often undiagnosed or misdiagnosed, but is an important cause of secondary infertility or secondary amenorrhoea. But with proper diagnosis and management this disorder can be treated.

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