

## Osseous Metaplasia of the Endometrium: A Rare Entity

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### About the Author



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### Introduction

This paper presents a case of osseous metaplasia of the endometrium, a rarely encountered disease. Metaplasia is described as a change of one epithelium into another epithelium due to some triggering factor. In this case, endometrium is converted into osseous spicules. It might be followed by an abortion presenting with Infertility or chronic endometritis or metabolic disorder like hypervitaminosis D or electroexcision of cervix, prolonged estrogen therapy, which is generally diagnosed by hysteroscopy and ultrasound and certified by histopathological examination.

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We report the case who presented with osseous metaplasia 17 years post-menopausal and 34-years post-abortion, the longest ever reported in the literature so far.

### Case Report

A 67-year-old female, P6L6A2, attained menopause 17 years ago and presented with complaints of lower abdominal pain with mucoid discharge per vagina on and off for 3 months. There was no history of per vaginal bleeding. No other associated symptoms like loose stools and vomiting were also reported. Bladder and bowel habits were normal. No significant past medical or surgical history was found. She is a P6L6A2. She had all normal vaginal deliveries, with last child birth 35 years ago. She had two abortions for which dilatation and curettage was done. Last abortion was 34 years ago. Her previous menstrual history had regular cycles, 2–4 days in 28–30 days, with moderate flow. She attained menopause 17 years ago and had no history of any post-menopausal bleeding.

Examination revealed normal general condition. Abdominal examination was normal, and no palpable mass was detected. External examination revealed normal external genitalia. Per speculum showed healthy cervix and dry vagina. Bimanual pelvic examination revealed atrophic uterus and free fornices.

Pap smear was negative for intraepithelial lesion or malignancy. Ultrasound done showed fluid-filled cavity or hydrometra measuring  $5.4 \times 4.5 \times 3.5$  cm with calcified material measuring  $1.9 \times 0.8$  cm in the cavity (Fig. 1). Hysteroscopy with biopsy was done using glycine as the medium. Cervical canal was normal. Anterior wall was normal. Right ostia and posterior wall were covered with bony spicules. Left ostia was seen (Fig. 2).

Biopsy of decalcified sections shows the trabeculae of a mineralized bone and inter-trabecular fatty tissue, negative for malignancy; mineralized bone trabecula represents Osseous metaplasia of endometrium. Endometrium revealed mucinous inflammatory exudates, negative for malignancy. Cervical biopsy was suggestive of chronic cervicitis (Fig. 1).

## Discussion

Endometrial osseous metaplasia is a rare clinical entity with the presence of mature or immature bone in the endometrium. It is an exogenous non neoplastic pathologic condition. Ossification can also be seen in cervix, ovary



**Fig. 2** Hysteroscopy image of Bony spicules in endometrial cavity

[1], and vagina [2]. Bhatia and Hoshiko reported a case of osseous metaplasia involving both the endometrium and the endocervix [3] in a 24-year-old woman.

Sorinala et al. [4] and Virchow attributed the formation of bone in the endometrium to spontaneous differentiation of fibroblasts into osteoblasts in two separate literatures in 1884. In 1956, De Brux et al. provided the first description of osteogenesis within the genital tract. In 1923, Thaler et al described the causal effect of osseous metaplasia to abortion.

Incidence is less than three cases per 10,000. The total number of cases throughout the world is <100 in the review of literature. Most of them are reported in post-menopausal women with abortions in the reproductive age groups and with infertility. A history of previous pregnancy is reported in > 80 % of the cases.

**Fig. 1** Ultrasound Image: Hydrometra with Irregular echogenic tissue



Pathogenic mechanisms related to the histogenesis of heterotopic bone into the endometrium are controversial. Osseous metaplasia from multipotential stromal cells, usually fibroblasts and mullerian derived cells which become osteoblasts by effect of irritants, inflammation or curettage [5]. Continuous and strong endometrial estrogenic stimulation is also one of the proposed cause [3]. Tuberculosis causes inflammation which acts as a promoter of secondary osteogenesis.

Ectopic bone formation and calcification result from the insult of chronic inflammation or tissue destruction with repeated abortions. Melius et al. [6] reported two cases of prolonged intrauterine retention of fetal bones following spontaneous abortions 13 years and 14 months before diagnosis. In the case reported by Ganem, some of the bone fragments in the endometrium contained marrow. Among the few reported cases in the literature, the time lag between antecedent abortion and discovery of the endometrial ossification varies between 8 weeks and 14 years.

Another proposed theory was chronic endometrial inflammation such as endometritis or pyometra. Bahceci and Demire [7] suggested that post-abortive chronic endometritis stimulates the release of superoxide radicals and tumor necrosis factor from the inflammatory cells. Metabolic disorders such as hypercalcemia, hypervitaminosis D or hyperphosphatemia are other causes. Cayuela et al. [8] studied DNA pattern and concluded that pluripotent mesenchymal cells, mullerian cells, and fibroblasts undergo osteoblastic metaplasia.

Clinical features include menstrual irregularities, pelvic pain, dysmenorrhoea, vaginal discharge, spontaneous elimination of bony fragments in menses, secondary infertility. Infertility occurs because bone is a foreign body acts as an IUCD, thus interfering with contraception. Its removal restores fertility [9]. The degree to which uterine cavity is involved is of particular clinical relevance.

Pathologists have to keep in mind the possibility of malignant mixed mullerian tumor of the endometrium during the diagnosis of osseous metaplasia of the endometrium [10]. Intrauterine contraceptive device, endometrial tuberculosis and retained foetal tissue are the common differential diagnoses for the bone in the uterus.

On ultrasound they usually appear like an intrauterine contraceptive device. In some cases these are visualised as filling defects on hystero-salpingography. Osseous lamellae which are white and either fan or disc shaped are reticulated, deeply embedded in mucosa and may have the appearance of flat coral.

Vigorous curettage should be avoided which may lead to synechiae formation. Recent studies recommend hysteroscopic removal of the bone under the ultrasonic guidance that helps proper visualization and complete removal of the bony spicules that may be embedded in the myometrium

[11], [12]. Moon et al described 10 spontaneous pregnancies following intrauterine bone removal in 11 patients.

Laparoscopic control during the procedure has been reported resulting in greater accuracy and prevention of complications such as uterine perforation. Sometimes hysterectomy is necessary for the definitive treatment of the patient.

## Conclusion

We consider that osseous metaplasia of the uterus in our patient was produced by the calcification of the endometrium during the healing process after an abortion. Our patient presented with Osseous Metaplasia 17 years after the onset of menopause and 34 years post abortion—the longest ever reported in literature

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