



## Novel Management of Endometrial Osseous Metaplasia

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### Abstract

**Background:** Osseous metaplasia of the endometrium is rare. Hysteroscopic resection of the bony fragments is usually offered. However, if the bony tissue is adherent or embedded in the myometrium, there is a risk of uterine perforation when the resectoscope or hysteroscopic scissors are used.

**Case:** A 37 year old woman, presenting with secondary subfertility, was diagnosed with osseous metaplasia based on ultrasound and hysteroscopy. We report a novel technique undertaken jointly with urology team. Under general anesthesia, rigid operative hysteroscopy was performed and lithotripsy device was used to fragment the osseous tissue in the uterine cavity. The bony fragments were retrieved using hysteroscopic graspers.

**Conclusion:** Per Cervical Utero Lithotripsy is minimally invasive and effective, making it a safer option as compared to other interventions.

**Keywords:** Endometrial; Osseous metaplasia; Hysteroscopic

### Key Points

1. Endometrial osseous metaplasia can present with a history of menstrual problems or subfertility, although it can be an incidental finding in asymptomatic patients. Pelvic ultrasound scan and hysteroscopy are useful tools for early diagnosis.
2. Complete resection of osseous metaplasia restores fertility in the majority of cases.
3. Per-Cervical Utero Lithotripsy (PCUL) is a novel technique which can be used selectively in particularly large cases of endometrial osseous metaplasia and ameliorates the risk of perforation associated with hysteroscopic resection.

### Introduction

Osseous metaplasia is defined by the presence of heterotopic normal bone tissue in a soft tissue. Osseous metaplasia of endometrium is a rare condition and there is no recommended treatment strategy described in the literature. The traditional treatment modality followed is hysteroscopic resection of the bony fragments (Figure 1). However, if the bony tissue is adherent/embedded in the myometrium, there is a risk of uterine perforation when the resectoscope/hysteroscopic scissors are used. There have been case reports describing even hysterectomy as a treatment intervention in such cases. The novel technique of Per-Cervical Utero Lithotripsy is a minimally invasive and effective treatment modality which reduces these risks associated with treatment of osseous metaplasia.

### Case Presentation

A 37 year old woman was referred to our unit with oligomenorrhoea and a four-year history of secondary sub-fertility following a termination of pregnancy 10 years ago. She had undergone a diagnostic laparoscopy and hysteroscopy 3 years prior to this as part of routine investigations for sub-fertility elsewhere. The laparoscopy was unremarkable with bilateral patent tubes. A small area of calcification was documented at hysteroscopy, which was avulsed and removed at the time. The patient had a transvaginal ultrasound scan which identified two linear echogenic areas within the endometrial cavity measuring 17 mm and 3 mm in length with posterior wall enhancement in the mid-uterine cavity. An outpatient hysteroscopy revealed a large area of bony tissue occupying the entire endometrial cavity. A tentative diagnosis of osseous metaplasia was made. Hysteroscopy was undertaken under general anesthetic jointly with the urologists. The plan was to apply techniques undertaken for the percutaneous management of upper urinary tract stones and treat the bony

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**Figure 1:** Hysteroscopic finding of osseous metaplasia.



**Figure 2:** Part of specimen removed- Histology confirmed endometrial osseous metaplasia.

tissue in a similar manner. Prior to the hysteroscopy the patient underwent a CT pelvis to exclude myometrial invasion. Standard rigid saline hysteroscopy was performed prior to the insertion of a 24F nephroscope (Karl Storz), which was easily negotiated through the uterine cavity to identify the bony lesion. A Swiss Lithoclast Master (EMS) with dual ultrasonic and pneumatic lithoclast attachments was used to fragment the osseous tissue, which easily dislodged and floated freely within the cavity. The bony fragments were then retrieved successfully using hysteroscopic graspers. Histology confirmed endometrial osseous metaplasia (Figure 2). The patient proceeded with a cycle of IVF using an antagonist regime. Nine oocytes were collected, two fertilized and a single day 2 embryo was transferred, which failed to implant. No embryos were suitable for cryopreservation. A second look hysteroscopy was performed a year later and there was no residual calcification seen.

## Discussion

Osseous metaplasia occurs most commonly within the endometrium. However, cases have been reported involving the cervix, vagina, and ovaries [1-3]. A rare case of osseous metaplasia of ovarian thecoma along with endometrial adenocarcinoma can also be found in the literature [3].

Most of the cases are asymptomatic. Women can present with secondary sub-fertility [4], menstrual irregularities [5], abnormal vaginal bleeding, discharge, dysmenorrhoea and chronic pelvic pain [6]. Bony fragments have been identified as early as 8 weeks and up to 15 years after an antecedent pregnancy that ended in either miscarriage or termination. Rarely, it has been reported in perimenopausal women with menstrual problems. Most of the cases of

endometrial osseous metaplasia are diagnosed incidentally in view of its long asymptomatic phase. Hysteroscopy was the diagnostic investigation for the majority [4-6]. Various surgical treatments have been proposed. Hysteroscopic resection is the standard treatment. The resection is performed by diathermy, or loop resectoscope. The most serious complication of hysteroscopic resection is uterine perforation and its sequelae [1]. Laparoscopy may be subsequently required to exclude any such complication. Cases of hysterectomy have been reported as treatment for endometrial osseous metaplasia [7]. Using laser as an energy source seems a reasonable option if the resultant fragments are retrieved. The authors felt that the aforementioned PCUL technique allowed for a more reliable clearance of all the osseous metaplastic tissue and therefore reduced the risk of retained material. Conjugate oestrogen can be administered for 6 to 8 weeks following the resection to promote endometrium proliferation [5].

Endometrial osseous metaplasia may be incorrectly diagnosed as retained IUCD and calcification. It is important to differentiate it histologically from mixed müllerian tumors, in order to avoid an unnecessary hysterectomy [8]. Finally, uterine gliosis with ossification, a benign condition arising from osseous metaplasia of endometrial stroma (in most cases as a result of implantation of fetal tissues following instrumental abortion) should be distinguished from uterine teratomas and mixed mesodermal tumors in order to avoid an unnecessary hysterectomy [8].

In order to avoid hysterectomy as well as to minimize complications such as perforation, we involved urologists in the management of this case, given their familiarity with the equipment which is routinely used during percutaneous renal stone surgery (Per Cutaneous Nephro Lithotomy). Using the lithoclast, we managed to break the large bony tissue into smaller fragments, which could be easily removed hysteroscopically, thus minimizing the risk of perforation.

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