Video Case Report

Osseous Metaplasia of the Endometrium and Successful Hysteroscopic Resection: A Video Case and Review of the Literature

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Abstract

Osseous metaplasia of the endometrium is defined by the presence of mature or immature bone tissue in the endometrium. Most of the cases are associated with secondary infertility after abortion, chronic endometritis, or the presence of foreign bodies in the endometrium. Some cases are asymptomatic; others have menstrual abnormalities such as menorrhagia or oligomenorrhea. Osseous metaplasia is mostly seen after recurrent abortions. Removing the bone tissue helps spontaneous conception. Intrauterine hyperechogenic lesion, suggesting calcification in transvaginal ultrasonography, creates suspicion in diagnosis. Here, we present a patient who underwent dilatation and curettage procedure following a missed abortion, and osseous metaplasia of endometrium was radiologically detected at a 1-month follow-up examination. White bony material was shown in the uterine cavity with hysteroscopy. The lesion was treated by hysteroscopic removal without any complications. Histology confirmed the diagnosis of endometrial osseous metaplasia. Thus, hysteroscopy was effective in the diagnosis and treatment of endometrial osseous metaplasia.

Keywords: Endometrium, hysteroscopy, osseous metaplasia, resection

Introduction

Osseous metaplasia of the endometrium is a rare condition defined by the presence of ectopic bone tissue in the endometrium.[1] It has an estimated incidence of 3/10,000. So far, 100 cases have been reported in the literature, and in more than 80%, the condition was observed following a pregnancy.[2] Osseous metaplasia is most commonly associated with secondary infertility. Other symptoms include menstrual irregularities, pelvic pain, dyspareunia, and vaginal discharge. Most cases have a history of curettage.[3] Several pathophysiological theories have been proposed and the most accepted theory is metaplasia of stromal cells to osteoblastic cells that produce bone tissue. Sonographic examination, showing characteristic hyperechogenic bone tissue in the uterus, helps in the diagnosis. However, hysteroscopy is the gold standard. Complete removal of bony spines from the endometrial cavity with hysteroscopy under sonographic guidance is usually the cure of the lesion.[3]

Case Report

The patient who underwent curettage at 7 weeks of gestation, according to her past menstrual date at another center 1 month before her control appointment at our clinic presented without any symptoms or discomfort due to curettage. Before the

Article History:
Submitted: 2-Nov-2021
Revised: 31-Mar-2022
Accepted: 4-May-2022
Published: 7-Nov-2022

Access this article online

Quick Response Code:

Website: www.e-gmit.com

DOI: 10.4103/gmit.gmit_121_21

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How to cite this article: Kadirogullari P, Seckin KD, Selcuki NF. Osseous metaplasia of the endometrium and successful hysteroscopic resection: A video case and review of the literature. Gynecol Minim Invasive Ther 2022;11:244-6.

procedure, the patient's menstrual cycle was regular, and there was no history of endocrinological disorders, tuberculosis, or any other chronic illnesses. She did not receive an intrauterine device (IUD) for contraception following the curettage. Her physical examination was normal, and no pathology was observed during the bimanual pelvic examination. Her routine blood tests were normal.

Transvaginal ultrasound examination was performed using a Voluson E8 scanner and a 6–12 MHz transvaginal probe (GE, Healthcare). Longitudinal and transverse sonograms of the pelvis revealed well-defined, irregular, linear, echogenic areas within the endometrium, as well as in the upper part of the cervix along the endocervical canal, with posterior acoustic shadowing [Figure 1]. Myometrium and ectocervix were normal. The rest of the pelvic structures were also unremarkable. Based on sonographic findings and clinical history, the diagnosis of osseous metaplasia of endometrium was suspected.

These bony fragments were removed by hysteroscopy [Supplementary Video 1]. The patient was planned for operative hysteroscopy under total anesthesia. An oblique 30° optical instrument was used to visualize the intrauterine cavity. Saline was used as uterine distension media. Multiple bony spicules measuring 0.5–2 cm were visualized and removed using a hysteroscopic grasper. Then, the samples obtained were examined histopathologically. The H- and E-stained microphotographs [Figure 2] confirmed the presence of trabecular bone along with secretory endometrial glands, endocervical epithelium, and decidualized stroma at places. Cartilage, bone marrow, or trophoblastic tissue were not seen. Written informed consent was obtained from the patient for publication of this video article and any accompanying images.

Discussion

Osseous metaplasia of the endometrium is a rare entity with its unclear etiology and pathogenesis.[3] These intrauterine structures are generally not suspected clinically. They remain asymptomatic until they are seen on ultrasonographic examination. Osseous lamels are seen as white, fan-shaped, or disk-shaped structures embedded in the mucosa or as an intracavitary structure with hysteroscopy.[3]

Osseous metaplasia of the endometrium acts like an IUD, making the endometrium unsuitable for embryonic implantation. This hypothesis has been confirmed by cases of spontaneous pregnancies reported in the literature following the removal of these bone fragments.[1] This lesion may be misdiagnosed as an IUD due to its rarity and resemblance of an IUD under ultrasonography. Symptoms include pelvic pain and menstrual changes, but infertility is the main result of the presence of bone tissue in the uterine cavity.[1]

The lesion is typically bone tissue surrounded by normal endometrium on histopathologic examination. Two main theories have been proposed regarding the origin of the bone in the endometrium. According to one hypothesis, bone metaplasia develops as a result of embryonic bones that continues to develop after abortion.[4] The fetal bones retained after the second-trimester miscarriages can lead to ossification,[4] which causes chronic inflammation and tissue damage that results in osseous metaplasia.[5] Few cases of miscarriage have been reported in very early pregnancy without fetal bone tissue. The second theory suggests that bone metaplasia is caused by multipotent stromal cells, often fibroblasts that develop into osteoblasts.[5] This metaplastic transformation may be due to prolonged estrogenic stimulation, chronic endometrial inflammation such as endometritis or pyometra, and metabolic disorders such as

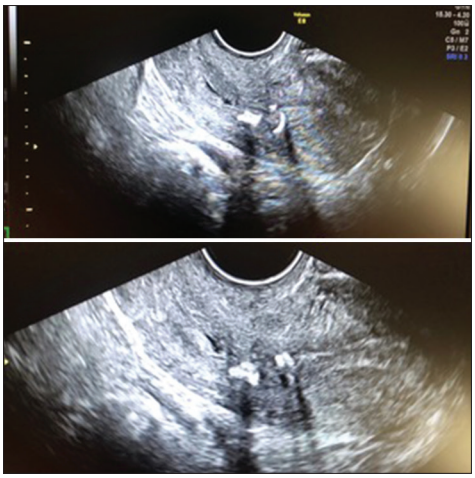

Figure 1: The appearance of hyperechogenic lesions in the uterine cavity through transvaginal sonography

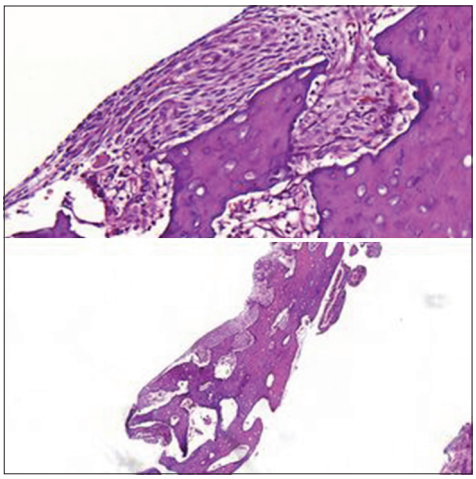

Figure 2: The histopathological seen of endometrial osseous metaplasia http://www.apagemit.com/page/video/show.aspx?num=278

hypercalcemia or hyperphosphatemia.[6] It may occur due to chronic inflammation caused by conditions of endometritis or chlamydia infection.[7]

It has also been suggested that postabortion chronic endometritis stimulates the release of superoxide radicals and tumor necrosis factors from inflammatory cells.[8] This leads to metaplasia of stromal cells to osteoblastic cells in women with insufficient superoxide dismutase activity. Metaplasia seen in nulliparous patients represents true osseous metaplasia as in the present case. Tulandi *et al*. reported a case in which they found that endometrial tissue was genetically derived from fetal tissue.[9] In contrast, Cayuela *et al*. reported a case of true osseous metaplasia in which DNA from ossified tissue was taken from the woman.[2] On the genetic analysis of the cause of bone metaplasia, Parente *et al*. also showed that DNA from metaplastic tissue has the same origin as the patient.[10] It has been reported that the time interval between abortion and bone metaplasia can range from 8 weeks to 23 years.[10] Van den Bosch *et al*. noted asymptomatic osseous metaplasia in the uterus 20 days after spontaneous delivery. Due to the short-time interval between birth and diagnosis and the histological features of the lesion, they concluded that bone metaplasia was caused by the first pregnancy terminated in the midtrimester and is already present in the second pregnancy.[6]

Diagnosis is made after hysteroscopic sampling and histopathological examination of the suspected lesion in ultrasonography. The main finding in the ultrasonographic examination is hyperechoic linear or irregular or irregular areas in the endometrium with posterior acoustic shadowing. There may also be involvement in the endocervical area. There may also be signs of unusual ossification in the cervix, ovaries, and vagina.[3]

The differential diagnosis includes the presence of IUD, foreign bodies, Asherman's syndrome, calcified submucosal fibrosis, and Müllerian tumor. Magnetic resonance imaging of the pelvis helps to confirm ultrasound findings. Treatment for this condition is the removal of bone fragments with hysteroscopic guidance for histopathological analysis. An alternative is dilatation and curettage.[2,3] However, curettage should be avoided due to the risk of synechia formation. Recent studies support hysteroscopic removal of bone with ultrasound guidance as the gold standard in management. In our case, hysteroscopy was effective and sufficient for the diagnosis and treatment of endometrial osseous metaplasia.

Conclusion

Although osseous metaplasia of the endometrium is a rare entity, it can have devastating results such as secondary infertility. Due to its nonspecific symptoms, diagnosis can be challenging. Accurate diagnosis is important because excision with hysteroscopy can help young patients with fertility wish to conceive. Therefore, osseous metaplasia should be among the differential diagnosis of patients, especially presenting with secondary infertility.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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