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ENDOMETRIAL OSSEOUS METAPLASIA: A REPORT OF TWO CASES AND BRIEF LITERATURE REVIEW

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ABSTRACT

Endometrial osseous metaplasia is an uncommon entity with the presence of bone in the endometrium. In most cases reported, ossification was followed by an abortion and patients presented with secondary infertility. Here, we present two cases; the first has secondary infertility after voluntary interruption of pregnancy and the second has been admitted for persistent vaginal bleeding after vacuum aspiration. In both cases, transvaginal ultrasonography showed an intrauterine hyperechogenic lesion. Hysteroscopy revealed multiple white spicules of bony material in the uterine cavity. These lesions have been treated by hysteroscopic removal.

KEYWORDS: Osseous metaplasia; transvaginal ultrasonography; hysteroscopy.

INTRODUCTION

Endometrial osseous metaplasia is a rare lesion characterized by the presence of mature or immature bone in the endometrium. Around 100 cases have been reported in the literature and most of them were followed by a previous history of abortion.[1] Through the experience of two cases and brief literature review, we discuss the best practices for the diagnosis and management of this rare entity.

Case presentation

Case 1

A 28 years old nulliparous woman, presented to gynecology outpatient department with the history of secondary infertility. She was married for 3 years. Past history revealed a voluntary interruption of pregnancy two years before. Following the event, her menstrual history was normal. The semen analysis of the husband was normal. Transvaginal ultrasonography showed an echogenic area in the uterine cavity. Diagnostic hysteroscopy was done using rigid hysteroscope. Endometrial cavity revealed multiple small, hard bony spicules, which were removed using hysteroscopic forceps and submitted for histopathological study. The pathologic report was endometrial osseous metaplasia **(Figure 1)**. Further evaluation could not be done as the patient was lost for follow up.

Case 2

A 31 years old multiparous woman, admitted for persistent vaginal bleeding following vacuum aspiration of incomplete abortion. She did not have any other significant complaints. On admission, the patient was afebrile, with normal vital signs. Transvaginal ultrasonography revealed an echogenic area in the uterine cavity measuring 1×1cm suggestive of retained products of conception. The patient was subjected for operative hysteroscopy. The hysteroscopic appearance was a coral-like white plaque, grasped with the loop of the resectoscope and successfully removed without complications. Histopathology of the removed lesion showed fragments of bone surrounded by normal endometrial tissue **(Figure 2)**. Patient was monitored during the follow-up period and she had spontaneous conception 12 months after the procedure.

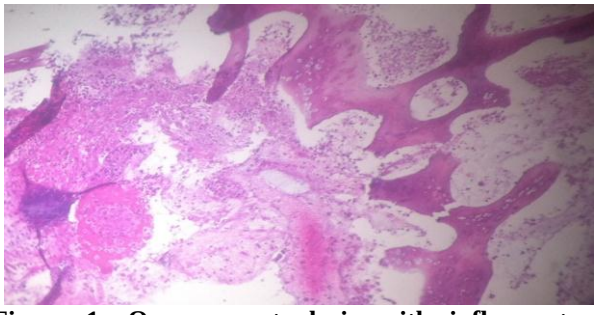

Figure 1: Osseous metaplasia with inflammatory reaction (HEx40)

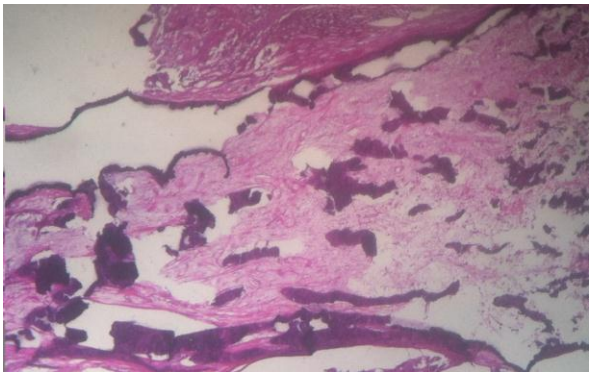

Figure 2: Osseous metaplasia with necrosis (HEx10)

DISCUSSION

Endometrial osseous metaplasia is an uncommon clinical entity with an estimated incidence of 3/10 000.[1] It is also described by various other names such as endometrial ossification and ectopic or heterotopic intrauterine bone.[2] Ossification is also reported in the cervix, the ovary and the vagina.[3,4,5] Like our cases, majority of the patients are in the reproductive age group with history of first trimester abortion, either therapeutic or spontaneous and have normal menstrual cycle in the post-abortive period as noted in case 1. However, a menstrual irregularity was noted in case 2. The time interval between the antecedent abortion and discovery of endometrial ossification varies from 8 weeks to 14 years.[6] Moreover, Shimazu described endometrial ossification in a 62 years old woman who also had the history of abortion 37 years earlier to the diagnosis.[7] Various theories have been proposed regarding the origin of bone in the endometrium. In fact, retained fetal bone after abortion, dystrophic calcifications of retained conceptual tissue, metaplastic transformation of fibroblasts into osteoblasts after long-term estrogenic stimulation or chronic endometrial inflammation and metabolic disorders such as hypercalcemia or hyperphosphatemia are the common proposed theories.[8,9] Recently, some studies regarding the genetic origin of tissue in osseous metaplasia have shown that DNA from the ossified tissue was derived from the woman supporting the osseous metaplasia theory.[1,10] The most common clinical presentation of osseous metaplasia is secondary infertility (like in the first patient), but it may also present with menstrual irregularities (like in the second patient), pelvic pain, dyspareunia, vaginal discharge, or an incidental finding.[11] The cause of infertility in women with this condition could be explained by osseous tissue acting as an intrauterine device. On ultrasonography, differential diagnoses of endometrial osseous metaplasia are endometrial tuberculosis, malignant mixed müllerian tumor and retained fetal tissue. It is important for the pathologists to distinguish this condition from the malignant müllerian tumor of the endometrium to avoid making a wrong diagnosis and hysterectomy in woman of reproductive age.[6,7] Hysteroscopy is considered to be the gold standard in diagnosis and management of endometrial osseous metaplasia, it appears as osseous lamellae, white in color, fan or disc shaped and embedded in the mucosa or as an intracavitary structure. In difficult cases with extensive osseous metaplasia, 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.[6,12] In both cases, hysteroscopy was effective in the diagnosis and treatment of endometrial osseous metaplasia without complications. The diagnosis is confirmed by histopathological examination of intrauterine materiel, objectifying osseous tissue surrounded by normal endometrium. The use of estrogen is controversial as it can promote osteogenesis and can be one of the causes of endometrial ossification.[13] In a woman with normal regular menstrual cycle, endogenous hormones are sufficient for endometrial regeneration.[6] Like in case 2, literature has shown good conception rates and pregnancy outcome after hysteroscopic removal of the ectopic intrauterine lesion in the most cases reported with secondary infertility. In case 1, with secondary infertility, further evaluation could not be done as the patient was lost for follow up.

CONCLUSION

Endometrial osseous metaplasia is a rare disorder that usually leads to secondary infertility and menstrual irregularities, especially following abortion. Hysteroscopic evacuation is the gold standard in diagnosis and management and most of the patients conceived after this procedure.

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