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Endometrial ossification: A case report

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Abstract

Endometrial ossification is uncommon condition which is related to secondary infertility following abortion. It is characterized by the presence of mature or metaplastic bone formation in the endometrium. There are about 100 cases that have been reported in the literature to date. Its etiology and pathogenesis is controversial and several debatable assumptions have been described in the literature. We present an additional case of endometrial ossification of a 32 years old female who presented with history of uterine Caesarian section delivery five years ago. She later had abortion of 14 months of gestation two years after Caesarian delivery. One year later after abortion, during pre conception examination, a mass was found in the uterus which was not treated until when she was referred to our hospital. Ultrasonography examination revealed a hyperechoic mass which size was 3.6×0.8cm, extending from lower uterine segment to endocervix. The mass was resected with uneventful recovery. The resected tissues were subjected for histological analysis where the diagnosis of endometrial ossification was made on histopathology which showed endometrial tissue intermingled with bony tissue.

Few months later she conceived and delivered normally a full tem healthy baby boy. The removal of bone fragments by curettage is very likely to bring about natural conception in patients with endometrial ossification.

Keywords: Endometrial ossification; Endometritis; Hysteroscopy

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Introduction

Endometrial ossification, synonymous to endometrial osseous metaplasia is a rare condition which is related to secondary infertility [1] commonly following one or several abortions. It is characterized by the presence of mature and immature bone tissue in the endometrium. It typically affects women in reproductive age group who had previous history of endometritis, spontenous, missed, incomplete or therapeutic abortion, or menstrual irregularities suggesting endochodral ossification. The patient with endometrial ossification may present with intrauterine pain, unexplained irregular vaginal bleeding, discharge or secondary infertility. A few cases occur without a history of neither pregnancy .nor secondary infertility.

The endometrium is known to have high differentiation ability in response to endogenous or exogenous hormonal stimulation or any reaction that may lead to inflammation of the endometrium. The reserve cells beneath endometrial glandular epithelial cells have a pluripontential to undergo metaplastic change of the paramesonephric duct. There are various theories that attempt to explain the presence of hard tissue in the endometrium. The most accepted theory is metaplastic transformation of endometral stromal cells and implanation of the stromal cells into osteoblastic cells that produce the bone. It is important to distinguish this condition from the mixed mullerian tumor of the endometrium to avoid hysterectomy. Removal of these bony bits leads to spontaneous conception. We present one such case in a 32-year-old female patient whose mass was diagnosed during pre-conception examination after experiencing abortion two years before.

Case History

A 32 year old female presented at Gynaecological department of First People's Hospital of Jining City with a history of uterine mass for two years by physical examination in another hospital. Her detailed medical history revealed that she had

undergone Cesarean delivery operation five years ago, and a voluntary induced abortion two years after Cesarean section. One year later after abortion, during pre-conception examination, an abnormal uterine mass was detected by B-mode ultrasonic diagnostic equipment which revealed a hyper-echoic mass in the uterine cavity. She was advised to attend specialized gynecological clinic for further investigations and management. However, no treatment was sought. Two years later after B-mode examination, the patient reported to the current hospital. She recounted normal menstruation cycles with moderate menstrual blood volume and no dysmenorrheal or abdominal pain. There was no lumbo-sacral regional soreness, irregular vaginal bleeding nor draining. Ultrasonography examination revealed a hyperechoic mass extending from lower uterine segment to endocervix. She was recommended for surgery and admitted. Further gynecological examination revealed normal vulva, smooth vagina and the cervix had mild erosion but no bleeding on contact. The uterus was located in normal anatomic position with normal size, irregular, moderate texture and no tenderness. There was no palpable abnormality. All vital signs and laboratory indices were within normal limits. The patient underwent a transurethral resection of intrauterine residue by hysteroscopy performed in the operating room. The weight of scrapings was five grams. The specimen was fixed with 4% formaldehyde solution, going through conventional paraffin embedding and slicing by 4um. After hematoxylin-eosin staining, the glass slices were observed under the microscope. Macroscopically the scrapings showed a pile of gray-red endometrioid broken tissue, mixed with gray osteoid tissue. The resected tissues were subjected for histological analysis. Where proliferative endometrium mixed with bone tissue with calcification was confirmed (Figure1-3). The pathological diagnosis of endometrial ossification was confirmed.

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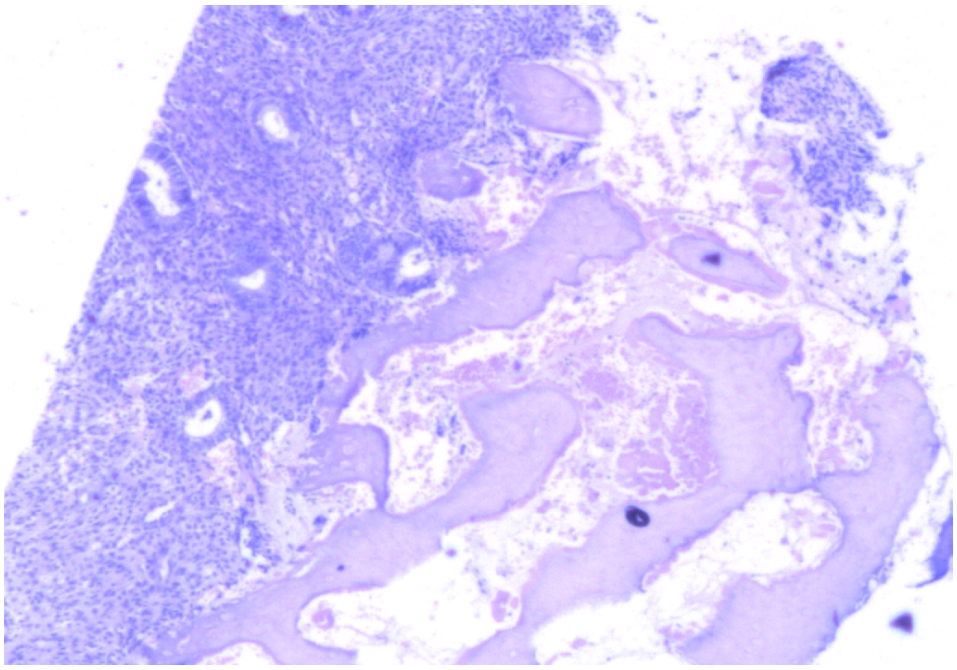

Figure1 Proliferative endometrium mixed with bone tissue with calcification

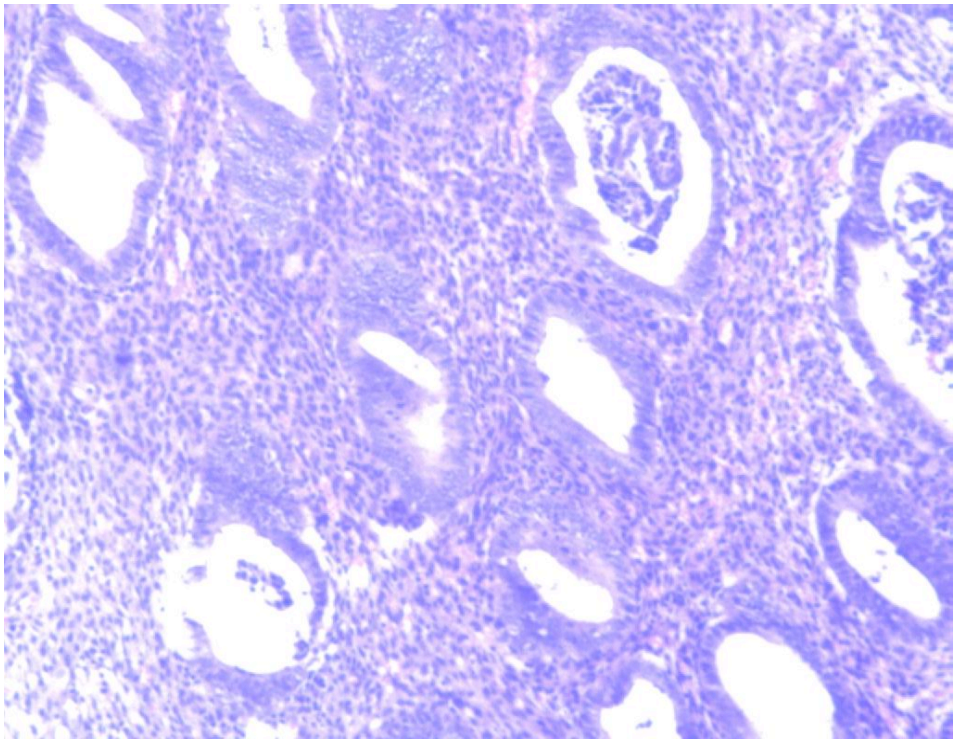

Figure 2 Proliferative endometrium

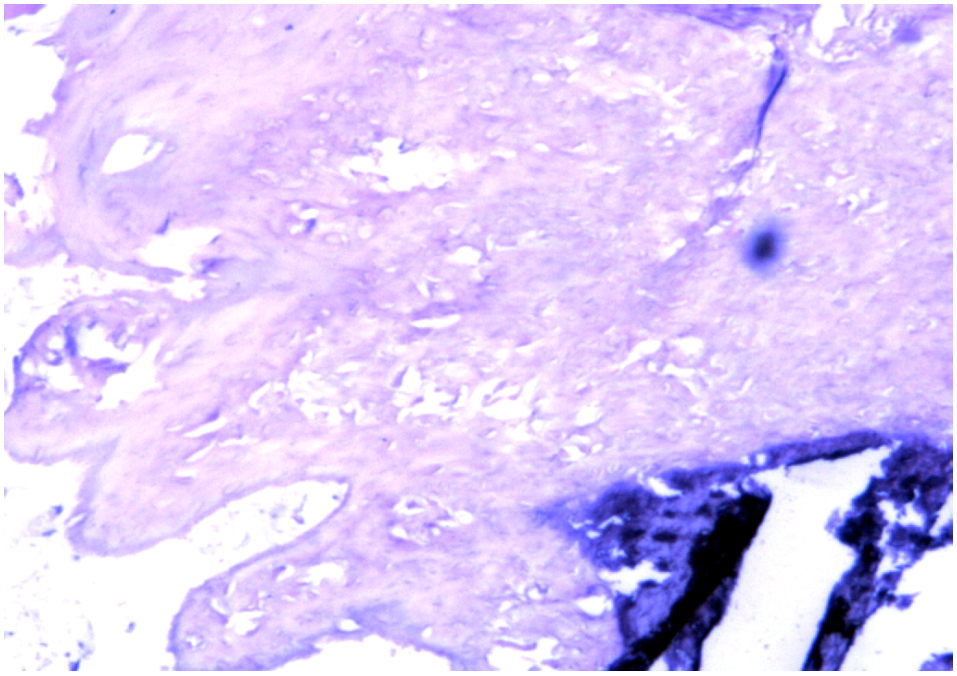

Figure 3. Bone tissue with calcification, no osteoblasts around trabeculae

Discussion

Endometrial ossification is rare pathological entity which with controversial etiology and pathogenesis. It is related to previous miscarriage or endometritis after abortion[2]; however, some scholars believe degeneration and necrosis of the embryonic residues to be the cause of metaplastic bone formation in the endometrium [3]. Other factors that have been implicated to be associated with pathogenesis of this condition include dystrophic calcification, abnormal alkaline phosphatase, metabolic disorders of calcium and phosphorus, and excessive estrogen stimulation. Although various scholars explain it differently, the most accepted the first two factors [4]. In our case, this patient had gone through previous miscarriage, so it was consistent with the first factor.

There are common clinical features that have been described in most literature. This disease occurs in women of childbearing from the age 20 to 40 years old. Most

patients have the history of recurrent spontaneous or induced abortion. A number of patients have reported experiencing fever after abortion which was accompanied with irregular vaginal bleeding, faint pain in the lower abdomen, and pus discharge per vagina. A set of patients have reported menstrual changes after abortion, such as menorrhagia, hypomenorrhea or dysmenorrheal. In few cases, vagina discharges with osteoid has been experienced [5]. Secondary infertility or habitual abortion has been encounterd in some cases and have been explained to be due to bone tissue from endometrial metaplasia acting as foreign body and therefore hindering embryo implantation. Very few of such patients may conceive and retain the pregnancy to term but the most will experience habitual abortion [6]。In the present case, the associating factor to the condition seems to be the induced abortion that occurred three years ago. Although there were no serious symptoms reported, the fact that the patient conceived immediately after treatment, the conception may have been hindered by the disease. Removal of bone material seems be one of the optional management of endometrial ossification particularly in order to conceive and deliver successfully.

B-mode ultrasonic diagnostic equipment has been very useful in preliminary diagnosis. The classical features that were also found in this case include several sizes of hyperechoic areas in the uterine wall or intrauterine cavity. Abdominal X-ray may show irregular dense shadow in the central pelvic cavity especially in severe cases. On the other hand, uterine lipiodol radiography is expected to show uterine cavity filling and/or defect or irregular shadows in the myometrium. Gray-white reflective enhanced tissue is visible in the hysteroscopy. And Ombelet W and other scholars had reported that transvaginal ultrasound and computed tomography could clearly show the endometrial pathology [7].

Morphologically, macroscopy shows a pile of grey-red endometrial tissue mixed with sizes and shapes of osteoid tissues, whereas microscopically, features of proliferative endometrium, secretory phase or menopausal changes are expected.

Chronic inflammatory response is commonly observered with infiltration of lymphocytes and plasma cells in the fibrotic endometrial stroma with ossification. The trabecular bone fragments are in different size and shape with in disorderly arrangement. Neither osteoblasts nor osteoclasts in the peripheral of the trabeculae. The trabecular bone can form lamellar bone in the endometrial stroma, and this characteristic is common in the chronic endometritis after abortion. If these trabecular bones were residue of embryonic bones, then they would be infantile and immature in nature and located in the endometrial surface, not in the endometrial stroma [8]. This supports that the pathogenesis of endometrial ossification is strongly associated with metaplastic changes rather than from residues of embryonic bones.

Repeated curettage and clearing the bone tissue in the uterine cavity is the best way to treat this condition. The curettage should be arranged during the three to five days after menstruation, because the endometrium is thin at this time, easy to take the bone and lead to less bleeding. The frequency is once a month, and the number of times depending on specific condition. Taking bone by the hysteroscopy in accurate position has very little damage, if necessary, one can get full cervical dilatation under anesthesia in order to clamp and scrape chunks of bone tissues. But for the older patient or if there have been a lot of bone tissue residues in the uterine cavity with serious infections implications , or when there is other gynecological surgical indications such as when one is suspicious of malignant mixed mullerian tumor, hysterectomy is recommended [9]. However, some scholars think that treatment of severe endometrial ossification may increase the risk of morbidly adherent placenta, presumably due to damage to endometrium, leading to abnormal placentation [10]. More clinical and experimental studies are recommended to validate the treatment options of this condition.

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