Bouzid et al., Gynecol Obstet (Sunnyvale) 2016, 6:3
http://dx.doi.org/10.4172/2161-0932.1000361

Gynecology & Obstetrics
ISSN: 2161-0932

Case Report Open Access

Osteoid Metaplasia of the Endometrium: A Case Report and Literature Review

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Abstract

Endometrial ossification is a rare condition. Most cases reported had a history of abortion. Clinical presentation may include abnormal vaginal bleeding or discharge, dysmenorrhea, pelvic pain and secondary infertility. Hysteroscopy appears to be the gold standard method to both diagnosis and treatment. We report a case of endometrial ossification in a woman who presented with pelvic pain. The patient had a pregnancy voluntarily terminated at an unknown gestational age. The diagnosis was suspected at Sonography. We insist on the fact that diagnosis must be considerate in a symptomatic or infertile woman with a history of both early and late abortion and illustrate the feasibility and safety of a hysteroscopic treatment for this condition.

Keywords: Endometrial ossification; Osseous metaplasia of the endometrium; Hysteroscopic resection

Introduction

Osseous metaplasia of the endometrium and cervix is an uncommon entity that causes infertility. This disorder is associated with the presence of bone in the uterine endometrium and occurs in approximately 0.3 per 1000 women [1]. In 1901, a German pathologist Mayer reported the presence of bone tissue in the uterus [2]. In 1923, Thaler [3] linked the presence of this bony tissue with a previous abortion. De Brux et al. [4] gave, in 1956, the first description of osteogenesis within the genital tract.

Various theories have been advanced for the etiology of osseous metaplasia of the endometrium [1]. We present a patient with osseous metaplasia, suspected on the sonogrphic examination and diagnosed and simultaneous treatment by hysteroscopy. We also review the literature.

Case Report

A 27 years old woman, presented to our institution complaining of dysmenorrhea and pelvic pain. She had one previous pregnancy which was voluntary terminated at age of 16. She had dilatation and curettage at an unknown gestational age. The patient had never undergone gynecological exam since that pregnancy. Clinical manifestations started one year earlier and the patient got married nine months ago.

Physical examination was unremarkable. Pelvic sonogram (revealed a regular size uterus with an endometrial echogenic region and posterior shadowing suggesting calcification, Figure 1).

Diagnostic hysteroscopy revealed two pieces of linear bone, first one near to the isthmus measuring 1 cm length, the second one in the corporeal measuring 2, 5 cm length extending to the lateral posterior uterine wall without reaching the uterine corne (Figure 2). The endometrium mucosa appeared to be inflammated. No other lesions were seen.

Surgical hysteroscopy removed the two fragments. Resectoscope was mainly used mechanically without electrosurgical energy to remove the fragment embedded into the endometrium. We also biopsied the endometrial mucosa.

Histological examination revealed mature bone tissue without osteoblastic activity mingled with proliferative endometrial glands and stroma site of nonspecific inflammation.

An antibiotic therapy was given. Pelvic sonogram performed one month after hysteroscopy revealed an irregular endometrium without ossification.

Discussion

Endometrial ossification is a rare disorder. The first case was reported over a century ago, but there still have been only a few cases reported up to day and most observations are case reports. It is an entity with controversial etiology and unclear pathogenesis.

Reported cases of endometrial ossification frequently have a history of previous pregnancy loss. It has been reported that the time interval between the abortion and osseous metaplasia can range from 8 weeks to 23 years [5,6].

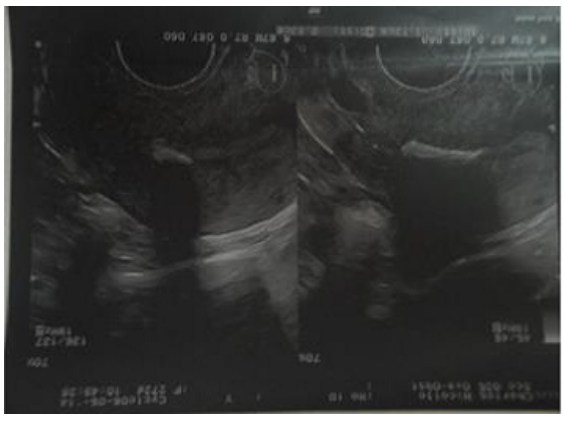

Figure 1: Ultrasound showing endometrial ossification.

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Received July 30, 2015; **Accepted** March 02, 2016; **Published** March 08, 2016

Citation: Bouzid A, Ayachi A, Mourali M (2016) Osteoid Metaplasia of the Endometrium: A Case Report and Literature Review. Gynecol Obstet (Sunnyvale) 6: 361. doi:10.4172/2161-0932.1000361

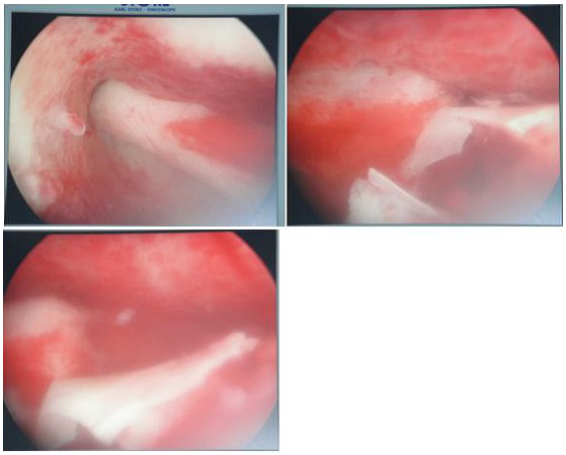

Figure 2: Diagnostic hysteroscopy revealed pieces of linear bone extending to the lateral posterior uterine wall. The endometrium mucosa appeared to be inflamed.

Symptomatology consist more often on menometrorrhagia, dysmenorrhea, vaginal discharge, pelvic pain, and spontaneous elimination of bony fragments in the menses.

Osseous metaplasia can also cause sub fertility by changing the milieu of the uterine cavity through the increase of production of prostaglandins.

Various theories have been advanced for the etiology of osseous metaplasia of the endometrium, including bone formation in the endometrium, spontaneous differentiation of fibroblasts into osteoblasts, direct implantation of fetal parts (in cases of second trimester abortions), repair process in response to chronic inflammation, and tissue destruction associated with repeated abortions [7].

The presence of retained fetal bones may lead to development of ossification, but this is valid for the second-trimester abortions [6]. However, for ossifications occurring after early abortions, osseous metaplasia of the endometrium is considered to be associated with chronic inflammation secondary to recurrent abortions and tissue damage [8].

Our patient did not remember the gestational age at the first abortion so we cannot consider an exact mechanism on our case.

It has been reported that endometrial ossification can also occur in the event of excess calcium or vitamin D consumption, in certain metabolic end endocrine diseases that lead to metastatic or heterotopic calcification, during long-term estrogenic stimulation of the endometrium, and in patients with a history of a hysterogram [9].

Ultrasonographic examination and confirmation by hysteroscopic examination are essential for the diagnosis of osseous metaplasia of the endometrium. Unlike the majority of cases reported, in the present case, osseous metaplasia was also determined in the cervix in addition to the uterus.

Many of previously reported patients were treated by hysterectomy or dilatation and curettage. However, both the young age of patients and the benign origin of this disorder recommend a more conservative treatment. Today, hysteroscopy is accepted to be the gold standard for the diagnostic and treatment. The most commonly cited hysteroscopic feature of this pathology is a white meshwork of bony spicules, frequently arising from the posterior wall and extending perpendicularly into the cavity, with a hard tactile consistency. Rarely, well-formed fetal bones, such as ribs, iliac bones, or humerus also may be encountered.

Hysteroscopic or resectoscopic excision of the bones is the mainstay of the treatment [2-11]. Ultrasonographic or laparoscopic guidance may be applied in extensive osseous metaplasias at which there may be deep myometrial invasion. However, it appears that the residual bones embedded within the myometrium do not appear to have a clinical significance.

Most patients with this condition presenting with infertility do so owing to the presence of a foreign body in the endometrium [12]. Spontaneous conception and birth are likely to occur when the osseous metaplasia of the endometrium is treated by the removal of bone fragments [13]. In our case, we intend to perform a second diagnostic hysteroscopy in case of absence of spontaneous pregnancy after 6 months.

The case reported here show no evidence favouring one pathogenic mechanism than another for endometrial ossification, we insist on the fact that it is a diagnosis considerate in a symptomatic or infertile woman with a history of both early and late abortion and illustrates the feasibility and safety of a hysteroscopic treatment for this condition.

Conclusion

Despite the high prevalence of abortions, osseous metaplasia of the endometrium is rarely encountered. This fact may be attributed to the misdiagnosis of the patients because of the nonspecific complaints or asymptomatic disease. Clinicians should keep this rare disorder in mind, especially in patients with a history of abortion.

Conflict of Interest

Authors declared they have no conflict of interest.

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