Rare disease

Osseous metaplasia of the endometrium

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Summary

Osseous metaplasia of the endometrium is very rare finding, and usually these cases presented with secondary infertility. Other symptoms are pelvic pain, dyspareunia, menstrual irregularities, vaginal discharge and the passage of bony fragments in menstrual blood. Suspicious lesion could see by hysterosalpingography or by ultrasonography; however, conclusive diagnosis and treatment tool is hysteroscopy. The aetiology is unknown, but theories include retained fetal bone and osseous metaplasia of endometrial tissue. We present a patient with osseous metaplasia who had treated with hysteroscopy.

BACKGROUND

This is a rare presentation of osseous metaplasia of the endometrium. The ultrasound images and hysteroscopic views were interesting.

CASE PRESENTATION

A 44-year-old woman with secondary subfertility was admitted to our infertility unit. She had had only one pregnancy 19 years before, and it had been terminated with

Figure 1 Sonography showing intracavitary calcification in the form of a densely echogenic band.

Figure 2 Osseous metaplasia of the endometrium as seen through hysteroscopy in our patient.

surgery at the second trimester. She had a regular menstrual cycle. She did not have abnormal vaginal bleeding or discharge, dysmenorrhoea, pelvic pain or dyspareunia. She did not have any gynaecological examination or treatment since abortion.

INVESTIGATIONS

Day 3 hormonal profile included the following parameters: E2:67, follicle-stimulating hormone: 7.8; luteinising hormone: 2.36. Investigation of the male partner revealed a variable sperm count of between 14 and 104 million/ml; however, he was found to have asthenoteratozoospermia. Transvaginal ultrasonography (TVUSG) examination on the third day of the cycle revealed a single, intensely echogenic, focus within the uterine cavity, which had an appearance similar to the intrauterine device (IUD) (figure 1). The right ovary had no antral follicles with normal appearance, and the left ovary had hypoechoic focus and no antral follicles.

DIFFERENTIAL DIAGNOSIS

Ultrasound examination revealed a single, intensely echogenic, focus within the uterine cavity, which had an appearance similar to the IUD.

TREATMENT

On diagnostic hysteroscopy, there were some adhesions at the internal ostium at the cervix. The adhesions were cut by office scissors. The hysteroscopic appearance was a thin, calcified, white, bony plaque within the cavity, which was grasped and removed from the cavity on operative hysteroscopy (figure 2). Endometrial curettage was also done. TVUSG confirmed that the uterus was empty.

Histological examination of the fragments removed from the uterus showed secretory endometrium with fragments of benign mature bone with marrow formation in the

stroma. Pathological examination revealed the diagnosis of osseous metaplasia of the endometrium (figure 3A, B).

OUTCOME AND FOLLOW-UP

The patient has an uneventful postoperative recovery, but elected to terminate infertility treatment because of the advanced age and decreased ovarian reserve, and she did not have spontaneous conception.

DISCUSSION

We report the case of a woman with osseous metaplasia who presented to our infertility clinic with secondary infertility after a previous termination of pregnancy. Despite the history of pregnancy termination, we suggest that osseous material is not secondary to the fetus because of the presence of bone marrow in the fragments removed from the cavity.

The aetiology of these bone fragments remains unknown. Two possible explanations are retention of fetal bone after an abortion that has occurred at ≥ 3 months gestation or osseous metaplasia of endometrial stroma. A possible mechanism of this metaplasia is that dystrophic calcification is a response to chronic inflammation, which then drives bone formation. This metaplastic change could be viewed as an adaptive substitution of one cell type for another that is better able to withstand an adverse environment. Another mechanism for osseous metaplasia is that it may have Mullerian origin, arising in the myoendometrial transitional zone. Metaplasias encountered in nulliparous patients must represent true osseous metaplasia, similar to that occurring after calcification of fibrosis or abscesses.

Bone is not populated with haematopoietic cells until the third trimester of pregnancy; thus, the presence of bone marrow in the fragments removed from the cavity as in this case can be considered as evidence against fetal origin. However, the presence of this marrow may be attributable

Figure 3 (A) Histopathologic examination of endometrial curettings showed bone and marrow formation (H&E $\times 100$). (B) The microphotograph reveals a well-circumscribed fragment of mature bone (arrows) surrounded by normal-appearing endometrium (H&E $\times 100$).

to bone being within the endometrial cavity for sufficient time to allow circulating haematopoietic stem cells to colonise the tissue and initiate marrow formation. This conflict between the two aetiologies could be resolved by DNA analysis of the fragments.

The presence of bony fragments within the endometrium may lead to subfertility by preventing implantation of a blastocyst, as with an IUD by increasing prostaglandin production, thus changing the milieu of the uterine cavity, or reactive endometritis caused by the fragments themselves may interfere with blastocysts implantation. Lewis *et al*¹ measured menstrual blood volume and PGE2 concentrations before and after the removal of retained fetal bones in a woman with infertility and menorrhagia. They found that the menstrual volume and total PG concentration decreased by 50% after the retained bone was removed.

Retained fetal bone will have symptoms of menometrorrhagia, dysmenorrhoea, vaginal discharge and pelvic pain, apart from subfertility.^{2 3} Sometimes these symptoms may not be diagnosed or their diagnosis can be too late. Differential diagnosis includes IUD, foreign bodies, calcified submucous fibroids and Ascherman's syndrome. In addition, endometrial ossification needs to be differentiated from mixed mesodermal tumours and adenosarcoma of the endometrium.⁴

Hysteroscopy remains the gold standard for diagnosis of endometrial pathology. The optimum treatment alternative for osseous metaplasia is hysteroscopy, as described by Frydman.⁵ Acharya *et al*⁶ claimed that they were the first to use the endoscopic resectoscope to diagnose and treat a woman with osseous metaplasia. Since then, there have been numerous reports in the literature on successful

hysteroscopy management of osseous metaplasia.^{3 7 8} If no other causes of infertility exist, pregnancy almost invariably follows their removal.^{7 9 10}

García León and Kably Ambe⁹ suggest that hysteroscopy is the first approach in the treatment and should be done with laparoscopy for the detection of other causes of infertility. Rodríguez and Adamson¹⁰ suggest that the hysteroscopic procedure should be viewed laparoscopically to reduce the risk of perforation. Coccia *et al*⁶ suggest that ultrasound guidance for hysteroscopic removal of extensive osseous metaplasia may represent a potentially safer and more effective alternative to laparoscopy and therefore encouraged further clinical evaluation of this technique. Elford and Claman¹¹ described a case of osseous metaplasia with an echogenic, but not hysteroscopic, appearance. It was treated with dilatation and curettage under USG guidance because there was deep penetration into the myometrium. Extensive or endocervical forms can generally be too deeply embedded to allow surgical hysteroscopic management.² Indeed, performing hysteroscopy at the same time as laparoscopy and dye insufflations can be disadvantageous in some cases. All cases must be evaluated with pre- or post-operative transvaginal ultrasound examination.

This case report confirms the importance of assessment of the endometrium in the initial investigation of patients with secondary infertility, particularly in those with a past history of late termination of pregnancy. The best practice for the diagnosis and treatment of osseous metaplasia is hysteroscopy. It allows complete removal of osseous fragments. Pathological examination should be done to confirm the diagnosis.

Learning points

- ▶ Retained fetal bone will have symptoms of menometrorrhagia, dysmenorrhoea, vaginal discharge and pelvic pain, apart from subfertility.^{2 3} Sometimes these symptoms may not be diagnosed or their diagnosis can be too late. Differential diagnosis includes IUD, foreign bodies, calcified submucous fibroids and Ascherman's syndrome. In addition, endometrial ossification needs to be differentiated from mixed mesodermal tumours and adenosarcoma of the endometrium.⁴
- ▶ Hysteroscopy remains the gold standard for diagnosis of endometrial pathology. The optimum treatment alternative for osseous metaplasia is hysteroscopy.

Competing interests None.

Patient consent Obtained.

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