CASE REPORT

Endometrial osseous metaplasia: an unusual cause of infertility

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SUMMARY

Osseous metaplasia of the endometrium is a rare disorder associated with the presence of bone in the uterine endometrium. Most patients with this condition presenting with infertility do so owing to the presence of a foreign body in the endometrium. We report a case of a 38-year-old woman who presented with secondary infertility due to osseous metaplasia in the endometrial cavity. She conceived spontaneously after hysteroscopic removal of the bony fragments from the uterus. Uterine osseous metaplasia is a rare cause of infertility that can be easily managed by hysteroscopic removal of the bony fragments, which results in return of fertility.

BACKGROUND

Osseous metaplasia of the endometrium is a rare disorder in which abnormal bone formation occurs in the endometrium. About 80 cases have been described in the literature.¹ The bone in the endometrial lining acts as an intrauterine device and results in infertility. Patients may also present with symptoms such as menstrual abnormalities, pelvic pain and dyspareunia. Most patients have a history of previous abortion. Hysteroscopic removal of the bony fragments usually results in resolution of symptoms and return of fertility.²

CASE PRESENTATION

A 38-year-old woman presented to us with a history of secondary infertility for 9 years. An earlier pregnancy had been voluntarily terminated at 20 weeks and was completed by a sharp curettage 9 years prior. The patient's only other complaint was that her husband felt pain with deep penetration during intercourse. Her menstrual

Figure 2 Hysteroscopy showing multiple bony fragments in the uterine cavity.

cycles were regular and she did not have any previous workup for the infertility. On speculum examination, a bony fragment was found to be protruding from the cervix.

INVESTIGATIONS

Transvaginal ultrasound revealed multiple linear calcifications in the uterine cavity ([figure 1](#)).

Hysteroscopy revealed multiple bony fragments in the uterine cavity ([figures 2 and 3](#); [video 1](#)).

TREATMENT

The fragments were removed using graspers inserted into the operating channel of a rigid hysteroscope. Since the fragments were not deeply embedded, hysteroscopic resection was not necessary in our patient. Postoperative sonogram revealed a normal endometrial lining ([figure 4](#)) and histopathology was osseous metaplasia of the endometrium.

Figure 1 Transvaginal ultrasound showing multiple linear calcifications in the uterine cavity.

Figure 3 Hysteroscopy showing bony fragments in the uterine cavity.

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Video 1 Hysteroscopic appearance of the bone in the uterine cavity.

OUTCOME AND FOLLOW-UP

The patient conceived spontaneously 8 months later, but the outcome was a missed abortion.

DISCUSSION

Osseous metaplasia of the endometrium is an uncommon condition characterised by abnormal bone formation in the uterine endometrium. Two cases of endometrial adenocarcinoma associated with osseous metaplasia have been reported. In such cases, the distinction of this entity from mixed mesodermal müllerian tumour is important.³

In the majority of cases, the osseous change is preceded by a previous abortion. The interval between the diagnosis of endometrial ossification and the antecedent abortion ranges from 8 weeks to 37 years. Patients usually present with secondary infertility but can also manifest other symptoms such as menstrual irregularities, dyspareunia, pelvic pain and vaginal discharge.⁴ Owing to the rarity of this condition and presence of calcification, it can be misdiagnosed as an intrauterine device.⁵ Complete removal of the bony fragments usually results in return of fertility.

Figure 4 Postoperative sonogram showing a normal endometrial lining.

Various theories have been proposed to explain the pathophysiology of uterine cavity ossification, including osseous metaplasia from multipotent stromal cells as a response to chronic inflammation, osseous differentiation from mesenchymal stem cells, secondary osteogenesis after retention of fetal bones, dystrophic calcification of retained necrotic tissues, and metabolic disorders of calcium and vitamin D.⁶ Cayuela *et al*⁷ compared the DNA of a patient with that in the osseous fragment extracted and demonstrated an identical genetic origin, proving that the entity was a true metaplasia. The differentiation of this condition from the in utero bone retention after an abortion is important.⁸ The absence of a surrounding tissue reaction and endochondral ossification helps to differentiate osseous metaplasia from retained fetal bony tissue. Presence of bone marrow in the bony fragments has been reported in osseous metaplasia.

These patients usually regain fertility once the bony fragments are removed. Hysteroscopic removal of the bony fragments is the safest approach and also confirms the complete removal under direct visualisation.²

Learning points

- ▶ Osseous metaplasia of the endometrium is an unusual cause for infertility.
- ▶ Most patients have a history of previous abortion.
- ▶ On ultrasound, the bony fragments may be confused with an intrauterine device; awareness of this condition will facilitate early diagnosis.
- ▶ Complete hysteroscopic removal of the bony fragments will result in return of fertility.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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