CASE REPORT

Osseous Metaplasia of Endometrium Managed with Oestrogens

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About the Author

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Case Report

A young lady presented to the gynaecology OPD with secondary infertility. She had been married for 8 years and had delivered a 2.4-kg term baby 18 months after her marriage. One and a half year after child birth, she underwent a first trimester elective MTP by suction evacuation. Following this, she was not able to conceive despite cohabitation for 4–5 years and also complained of scanty

flow for 18 months at presentation. There was no history of fever or anorexia, but she complained of late evening fatigue. There was no history of tuberculosis or possible exposure, and she had safe sexual behaviour. She had been treated as PID at a private clinic for vaginal discharge. She had undergone an HSG which showed a normal uterus, a beaded left tube and a bilateral spill from both fimbrial ends. A pelvic ultrasound scan showed dense linear echogenic foci with post-acoustic shadowing suggestive of calcification in the endometrial cavity and cervical canal but normal ovaries. Her calcium, phosphorus and vitamin D levels, hormonal profile including parathormone were found to be normal. Husband's semen analysis was also normal.

She was investigated for active tuberculosis and was administered doxycycline and metronidazole for pelvic inflammatory disease. Her ESR was 16 mm, hemogram

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and an X-ray chest were normal, and mantoux was 12 mm by 12 mm. A pre-menstrual endometrial biopsy (EB) was negative for mycobacteria PCR and AFB and histopathology revealed early secretory endometrium with osseous metaplasia. A post-menstrual hystero-laparoscopy was done. On hysteroscopy, the endometrium appeared polypoidal, with flimsy thread-like structures in the form of a web with enmeshed tiny bony spicules over left cornual opening which were broken with a 5F ureteric catheter introduced through operative hysteroscope. One triangular piece of bony tissue approximately 1 cm size was removed from the endometrial cavity and sent for histopathological examination (HPE). Laparoscopy revealed bilateral convoluted and beaded tubes with minimal adhesions but prompt bilateral spill of dye from both fimbrial ends on chromotubation. The cervical canal had no evidence of any disease. Patient was discharged on advice to follow a fertile period to conceive. HPE of EB revealed proliferative endometrium with osseous metaplasia. No polyps were identified. The patient's menstrual flow normalized after the procedure.

Two years later, the patient returned with a recurrence of decreased menstrual flow and failure to conceive. A transvaginal ultrasound (TVS) was again suggestive of calcification in the endometrial cavity. A premenstrual hysteroscopy was done, and several fragile, calcified pieces of varying sizes were removed under vision (Fig. 1). The calcified pieces were 1–2 cm in size (Fig. 2) and were loosely attached to the endometrium on the left lateral wall by thin mesh-like structures. Fibrosis could be seen in the fundus. Besides the large pieces, there were tiny, white specks of calcification on left lateral wall and near left cornu. Both cornua as well as the endocervical canal were otherwise normal. Endometrial biopsy (EB) was performed

Fig. 2 Hysteroscopic removal of bone

and sent for histopathological examination (HPE) along with repeat TB-PCR and AFB culture, considering high incidence of tuberculosis in India. TB-PCR and AFB culture were negative but HPE revealed osseous metaplasia, acute on chronic inflammatory infiltrates, foreign body giant cell reaction and a fragment of early secretory endometrium (Fig. 3). The menstrual flow volume normalized following removal of calcified pieces.

A review of literature revealed around 100 reported cases. There is no definitive treatment for the condition. The only treatment suggested by all the authors is removal of the bone pieces. This strategy had not worked for our patient.

In the hope of preferentially stimulating the normal endometrial cells over the metaplastic cells, this patient was administered ethinyl estradiol 50 µg from day 5 to 25

Fig. 1 Hysteroscopic removal of bone pieces using biopsy forceps

Fig. 3 Histopathological microphotograph showing osseous metaplasia and acute on chronic inflammatory infiltrate

along with Medroxy progesterone acetate (MPA) 10 mg from day 16 to 25. After the first and the second cycles, patient had heavy withdrawal bleeding, due to which dose of oestrogen was reduced to 20 µg in the third cycle. A trans-vaginal ultrasound done after three cycles revealed an 11.3 mm endometrium without any calcification. The patient was administered only folic acid and left to follow fertile periods. She conceived within the first cycle and delivered a healthy 3 kg baby by LSCS done for obstetric indications. On her next visit 10 months after the LSCS, she was in lactational amenorrhoea and had a fresh trans-vaginal sonogram showing endometrial calcification. She is now desirous of another pregnancy.

Discussion

Osseous metaplasia is defined by the presence of heterotopic bone tissue in a soft tissue. Osseous metaplasia of endometrium is clinically diagnosed by presence of bone in the endometrium that mimics an IUCD on sonogram. Most reported patients presented with heavy menstrual bleeding, secondary infertility, pelvic pain or dyspareunia. However, our patient presented with scanty menstrual flow on both the occasions with secondary infertility. Majority of patients reported had a history of first or second trimester spontaneous or induced abortion followed by surgical intervention in form of curettage. Incidentally, most case reports are from developing countries. Besides the endometrium [1–4], osseous metaplasia has also been reported in cervix, ovary, mesosalpinx, lung, breast, skin and kidney allograft.

Pathophysiology of metaplastic transformation of endometrium is largely unknown. Various theories have been proposed, but they are yet to be substantiated. Deficient superoxide dismutase activity in endometrium leading to metaplasia of stromal cells has been suggested [5]. As oestrogens promote osteogenesis, it has been implicated as a causative factor and its use in treatment of osseous metaplasia contraindicated [6]. It has been suggested that removal of bony spicules from endometrium is enough to restore fertility. However, very few studies have mentioned a follow-up till conception. Repeated evacuation of the bony pieces may be required till pregnancy occurs. This has been done by various methods such as blind dilatation and curettage, removal with biopsy forceps via operative hysteroscope, resectoscope using cautery or laser energy and hysterectomy. Our experience of removing the bony pieces with the help of operative hysteroscope was highly satisfactory; it could be achieved successfully under local anaesthesia with minimal damage to the endometrium.

Unfortunately, our patient did not conceive after the first treatment and returned with similar complaints after 2 years, when her disease had worsened. In addition, there was appearance of acute and chronic inflammatory cells in the endometrium. Since removal of bony spicules does not alter the presence of osteocytes in endometrium, it is expected that the disease will recur. Infertility will persist unless conception occurs soon after the evacuation as happened in our case although the disease was mild at the time. It would be precocious to say that the oestrogen therapy given after the second evacuation helped in conception by rapid preparation of the endometrium, but it can safely be said that it did not worsen her prognosis, as it resulted in a successful pregnancy.

Research is required to ascertain if oestrogens stimulate metaplastic osteocytes. Larger trials will be required to establish role of oestrogens in treatment of osseous metaplasia of endometrium. A better understanding of aetiopathogenesis that triggers the metaplastic transformation is also required to prevent recurrences of the condition.

Compliance with Ethical Standards

Conflict of interest None.

Ethical Statement The article is submitted herewith after informed consent of the patient and proper permission from her. She was administered oestrogens for her condition with informed consent explaining the pros and cons. There is no use of experimental diagnostic procedures, experimental drugs or animal use involved in the case.

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