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Review Article

Retained Intrauterine Foetal Bones: A Review of Literature

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ABSTRACT

Retained bone fragments or intrauterine ossification is becoming increasingly common now a days. One of the reasons is increasing cases of illegal abortion. These fragments in the uterine cavity act as inadvertent contraceptives leading to secondary infertility. It is usually preceded by a history of abortion, either spontaneous or induced.

We present a series of 8 cases of intrauterine retention of fetal bones. Along with this we review the literature on intrauterine ossification diagnosed on hysteroscopy. Most of the patients presented with infertility or irregular cycles. After hysteroscopic removal of bone fragments all of them were relieved of their symptoms.

We reiterate the importance of taking a thorough history and the importance of transvaginal ultrasound and hysteroscopy in women with secondary infertility, highlighting on the presence of foreign body in uterine cavity acting as contraceptive. The authors review the literature on increasing occurrence and cause of uterine ossifications.

Keywords: Bone, Infertility, Hysteroscopy, Ultrasonography.

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INTRODUCTION

Worldwide records of 1995, shows that about 26 million legal and 20 million illegal abortions take place every year.[1] Almost all unsafe abortions (97%) are in developing countries, and over half (55%) are in Asia (mostly in south-central Asia).[2] Reliable data for the prevalence of unsafe abortion is generally not available, especially in countries where access to abortion is legally restricted. Total legal abortions reported in India In year 2012 were 661353.[3] All recent and large-scale studies have shown that legally performed abortions are extremely safe with complication rate of 0 to 0.7 per 100,000. The overall early complication rate due to hemorrhage, uterine perforation or cervical injury is between 0.01% and 1.16%.[4] Complications due to previous abortions such as Asherman's syndrome may occur. Since 50% of abortions are performed on young patients, these women may develop future fertility problems caused by previous procedures. We describe a case series of patients with secondary infertility and menstrual irregularity due to prolonged retention of intrauterine bone after a dilatation and curettage. A number of case reports have been published on the prolonged retention of fetal bones up to 23 years after an abortion, either spontaneous or induced.5 Presenting complaints of patients are dysmenorrhea, dysfunctional uterine bleeding, pelvic pain, dyspareunia, vaginal discharge or spontaneous passage of fetal bones.[6] Intrauterine bone fragments may result from retained fetal bones or osseous metaplasia or both. Cases can be discovered by vaginal ultrasound examination,[7,8] hysterosalpingography[9] and hysteroscopy.10 In our cases, the patients had complaint of either infertility or irregular cycles. Hysteroscopy was performed for all which showed retained bone fragments.

CASE DISCUSSION

Case 1

A 29-year-old woman P1L1A1 with previous LSCS 9 years back with a termination 8 years back at 12 weeks gestation presented with secondary infertility. Fertility work-up showed a regular menstrual cycle, with no biphasic temperature curve. The semen analysis of the male partner was normospermic. Transvaginal ultrasound revealed a normal-sized uterus, with an echogenic scarred endometrium. Hysterosalpingography showed no abnormalities. During her 1st hysteroscopy eleswhere, Asherman syndrome was found and treated with adhesiolysis. The patient was treated with six cycles of clomiphene citrate with IUI. A second hysteroscopy was performed by us, during which irregular bone fragments were removed. (Figure1) Two months later, ultrasound showed a normal empty uterine cavity with a normal endometrial lining. A repeat Hysterosalpingography showed no abnormalities. In six months the patient became pregnant spontaneously. A healthy live baby was delivered by LSCS.

Case 2

A 31-year-old woman presented to us with complaint of secondary infertility. She had history of 2 abortions, one 5 years back and second 3 yrs back. She also had 1 healthy female child
6 years old. Her Fertility work up showed regular cycle with husband semen analysis being normal. Her ultrasound showed calcification in the uterine cavity. We did a diagnostic and operative hysteroscopy for her. Her cavity on hysteroscopy showed multiple pieces of bone. (Figure 2) They were removed. Post-operative ultrasound and HSG after 1 week were normal. Patient was advised for conception from next cycle.

Case 3

A 29-year-old woman presented with complaints of irregular continuous bleeding per vaginum after abortion for 5 months. She had an abortion at 10 weeks gestation 5 months back. She had a healthy child 3 years old. Ultrasound showed retained products of conception. We did a diagnostic and operative hysteroscopy for her. Her cavity on hysteroscopy showed multiple chips of bone. (Figure 3) They were removed. Post-operative ultrasound was normal. She was relieved of her menstrual complaints.

Case 4

A 31-year-old woman mother of 4 children presented to us with complaint of polymenorrhagia. She had history of 2 abortions in past 1.5 years. Her ultrasound was inconclusive. We planned diagnostic and operative hysteroscopy for her. She had moderate degree of asherman syndrome with multiple large pieces of bone inside (Figure 4). Fetal long bones could be clearly made out. After the removal patient was put on contraceptive pills and relieved of her symptoms.

Case 5

A 53-year-old woman was planned for IVF due to secondary infertility came to us for pre IVF hysteroscopy. There was an unfortunate demise of her 19 year old child because was which she wanted to conceive again. She had history of 4 abortions in past 12 years. On hysteroscopy we found small chips of calcification embedded in the endometrium (Figure 5). After removal of those chips she was taken up for IVF. After 2 cycles she successfully conceived.

Case 6

A 33 year old came with complaints of oligomenorrhea. She had 2 healthy children 5years and 7 years old. She had history of 3 abortions in last 5 years. Her ultrasound showed intramural calcification. We did diagnostic and operative hysteroscopy for her and found severe degree of asherman syndrome with 1 large piece of bone (Figure 6). After removal she was advised contraception.

Case 7

A 28-year-old came with complaint of secondary infertility. She had 1 healthy baby of 3 years. She had history of 1 abortion 2 years back. Her ultrasound was unremarkable. On hysteroscopy multiple bone pieces were seen and removed. Presence of bones was confirmed on histopathology also (Figure 7). She successfully conceived after 6 months (Figure 8).

Case 8

A 22-year-old came with complaints of primary infertility for 2 years. Her fertility work up was normal. Her ultrasound showed irregular cavity. On hysteroscopy to our surprise multiple large pieces of bone were seen and removed. Later history of premarital conception and abortion 4 years ago was elicited. She successfully conceived after 4 months.

REVIEW OF LITERATURE

Many cases are reported in the literature of retained bone fragments preceding abortions. The data on it has been constantly reviewed again and again in time. Theories have been postulated on the cause of these bone fragments or intrauterine ossifications. Adamson and Sommers.[11] reported a case of endometrial ossification in a patient who was taking high dose of calcium and vitamin D for long term. Most of the cases reported in literature do not have any evidence of hypercalcemia or the conditions leading to hypercalcemia to support the theory of metastatic calcification. Roth and Taylor.[12] gave a theory that without a previous pregnancy loss, metaplasia of mature endometrial stromal cells into bony tissue can occur in response to chronic inflammation or trauma. Sporadic cases in past without antecedent pregnancy support this theory.[12-15] Over the past few years there are many case reports describing endometrial ossification.[16-20]

Chronic endometritis can stimulate the proliferation of mesenchymal cells with inherent property of metaplasia and can differentiate them into chondroblasts or osteoblasts.14 Shroff et al.[14] described a case of endometrial ossification in a woman presenting with primary infertility.

Melius et al.[21] reviewed 50 cases in the literature, out of which 80% occurred after pregnancy. Most cases of endometrial ossification are discovered because of prolonged secondary infertility after a late therapeutic abortion. Bhatia and Hoshiko22 incidentally on hysteroscopy found endometrial and cervical osseous metaplasia in a 24-year-old woman.

Heterotopia, dystrophic calcifications, and ossification of post-abortive endometritis, metastatic calcification, metaplasia in healing tissue, prolonged estrogenic therapy after abortion, and retained fetal bone are the common proposed theories.[16,19,22,23,24] Bahceci and Demirel[25] also said that post-abortive chronic endometritis stimulates the release of superoxide radicals and tumor necrosis factor from the inflammatory cells. Long-term exposure of superoxide radicals and tumor necrosis factor on multipotent stromal cells in patients with deficient superoxide dismutase activity in the endometrium leads to metaplasia of the stromal cells into osteoblastic cells. Cayuela et al.[26] studied DNA pattern in a 27-year-old woman with endometrial osseous metaplasia following a first trimester abortion. They found matching DNA pattern in the blood of the patient and the bone pieces removed from the endometrium. No genetic material of male or fetus was found. They concluded that pluripotent mesenchymal cells, müllerian cells, and fibroblasts can undergo osteoblastic metaplasia in response to inflammation or curettage. This strongly supports the osseous metaplasia theory. Even after multiple case reports and studies retained fetal bones is the most evident and acceptable theory. In all our cases of infertility there was a history of preceding abortion leading to retained fetal bones supporting the theory of retained bone fragments. Recently, it has been suggested that the incidence of this complication after induced or spontaneous abortions is underestimated in the literature.[13]

Most Common clinical manifestations are menstrual irregularities, pelvic pain, dyspareunia, vaginal discharge, and secondary infertility. Majority of the patients are of reproductive age group with history of antecedent abortion either therapeutic or spontaneous and have normal menstrual cycle in the post-abortive period as noted in few of our cases. The time interval between the abortion and diagnosis of endometrial ossification can varies from 8 weeks to 37 years as cited in the literature.[25] Shimazu and Nakayama[27] described endometrial ossification in a 62-year-old post-menopausal woman who also had the history of abortion 37 years earlier to the diagnosis of endometrial ossification. Old studies recommended a series of dilatation and curettage to removal of the bone fragments. They also suggested that a vigorous single curettage should be avoided as it may lead to synechiae formation.[22]

Fig 1: Pieces of fetal bones taken out during hysteroscopy in case 1.

Fig 2: Multiple bone fragments seen in uterine cavity in Case 2

Fig 3: Multiple bone fragments floating in the cavity in case 3

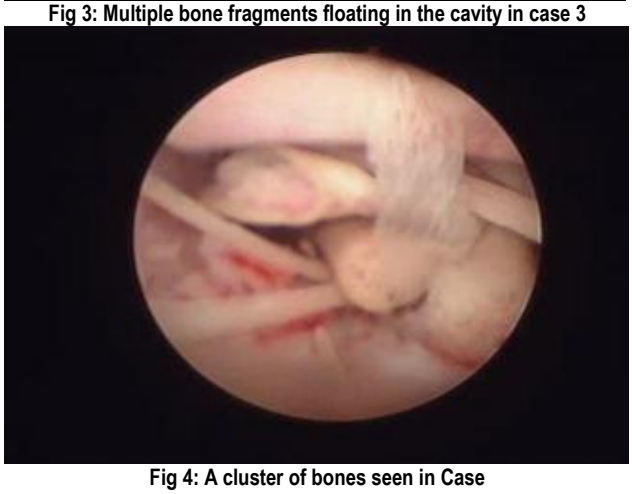

Fig 4: A cluster of bones seen in Case

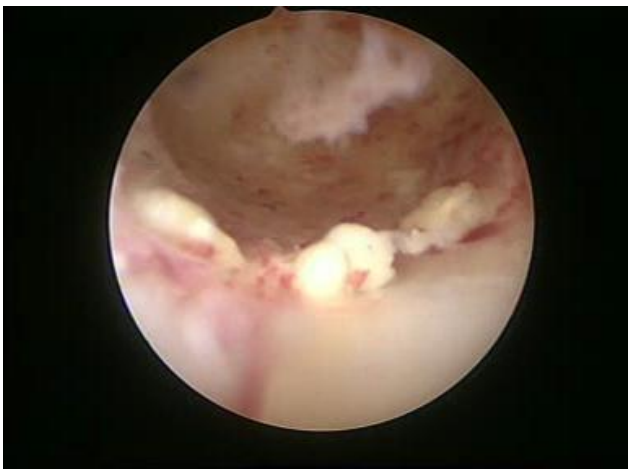

Fig 5: Small chips of impacted bone fragments in the posterior wall.

Fig 6: Large piece of bone seen in the cavity in case 6

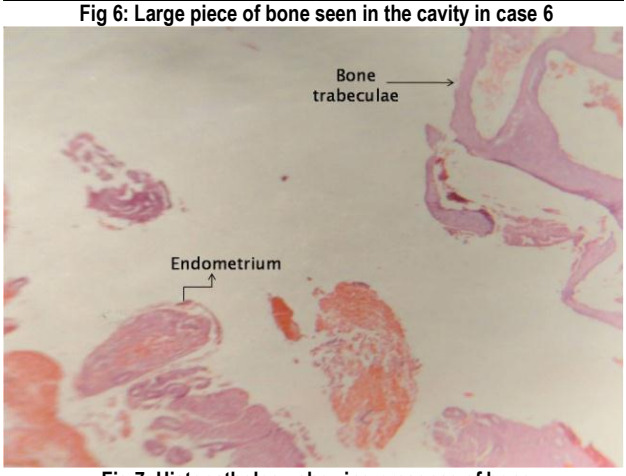

Fig 7: Histopathology showing presence of bone

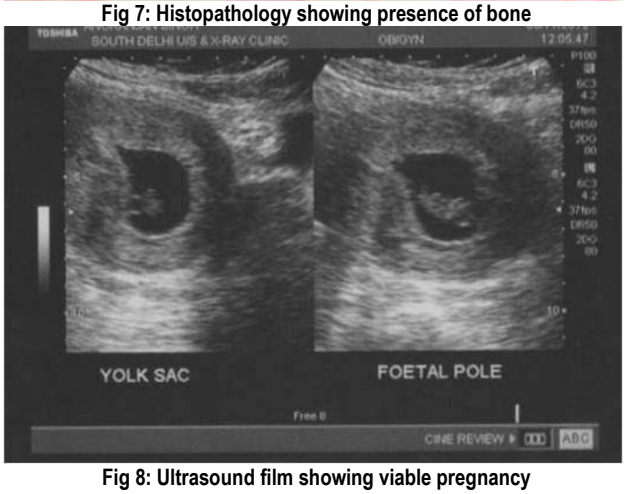

Fig 8: Ultrasound film showing viable pregnancy

Recent studies now recommend hysteroscopic removal of the bone with or without ultrasonic guidance as it helps in proper visualization and complete removal of the bony spicules that may be embedded in the myometrium.[25,26,28]

Bone in the endometrium can act as an intrauterine contraceptive device and its complete removal can restore the fertility and spontaneous conception.[16,24,25]

DISCUSSION

Every year many abortions are performed. Although abortion is generally a safe procedure but complications do occur. These cases reinstate an uncommon cause of secondary infertility or irregular cycles due to the prolonged retention of fetal bones where the patient had no complaints of pain, vaginal bleeding or discharge. Furthermore, it shows that fertility can be increased following removal of the fetal bones: generally, within 6 months of the hysteroscopy and removal of the bone fragments, the patients became pregnant. This suggests that the presence of fetal bone in the uterine cavity may act as an IUCD (5-9) by simulating IUCD like reaction. The diagnosis of retained fetal bone can be made by identifying a filling defect on a hysterosalpingogram, an echogenic area or calcification on vaginal ultrasound or, as in our cases, by direct visualization on hysteroscopy. Deeper impacted bone fragments, in the endometrium, on hysteroscopy, needs to be dissected and removed. A reason for suspicion should be an echogenic area in the endometrium on transvaginal ultrasound.[7]

CONCLUSION

We recommend a transvaginal ultrasound and hysteroscopy on every patient with a history of secondary infertility or abnormal cycles following abortion, regardless of the interval between the preceding termination and presentation. Taking a thorough history is a prerequisite. A high fertility success rate may be expected following the removal of any retained bones.

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