# Prolonged Intrauterine Fetal Bone Retention Following a Neglected Pregnancy: A Case Report

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**Abstract**- We describe a case of prolonged intrauterine fetal bone retention following a neglected pregnancy. A 43-year-old rural woman had one year history of malodorous vaginal discharge. She underwent medical treatment with broad spectrum antibiotics. Four days following the onset of antibiotic therapy, cystoscopy and hysteroscopy were done. Incredibly, some bony-like pieces were extracted by instrument from endocervical canal. Intrauterine retention of fetal bones, although rare, should be considered as a possibility in cases with unexplained abdominal pain, dysmenorrhea, dyspareunia, and long period infertility. Hysteroscopy is the best modality for diagnosis and may be enough for management of the condition in many cases. However, in complicated cases, hysterectomy would be inevitable.

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## Introduction

Intrauterine fetal bone retention is a rare condition with an estimated incidence of 0.15-0.26% per diagnostic hysteroscopies (1,2). After 13<sup>th</sup> week of gestation, bones start to form in embryo. If a neglected miscarriage or abortion happens after this time, uterus cannot absorb the bones, hence bones remain in uterine cavity and cause complications such as secondary infertility, chronic inflammation, vaginal discharge, dysmenorrhea, dyspareunia, menstrual irregularities, and pelvic pain (3). Herein, we describe a case of prolonged intrauterine fetal bone retention following a neglected pregnancy.

### **Case Report**

A G5L4A1 43-year-old rural woman, from one of the villages of Lordegan City in Chaharmahal and Bakhtiari Province, Iran, was referred to our center with one year history of malodorous vaginal discharge. According to the patient's history, her last pregnancy was about 10

years ago, and had terminated with missed abortion. Her other four previous pregnancies had terminated with live births via normal vaginal delivery. Following the last pregnancy, she had every-3-month-depo-medroxy progesterone injection as birth control method. Since then, she had long periods of amenorrhea interrupted by occasional spotting. On physical examination, abdomen was soft and non-tender and uterus seemed normal-sized. Vaginal examination showed no leakage from vagina itself; however, there was a malodorous watery discharge with air bubbles from external cervical os. Due to her unreliable menstrual history, pregnancy test was done which was negative. Uterine ultrasound study revealed some calcified linear foci within uterine cavity. On plain abdominal X-ray, some bones and air bubbles were seen in pelvic region. The possibility of fistula between uterus and adjacent pelvic organs was considered for the patient. On abdominopelvic computed tomography (CT) scan, a complex intrauterine mass was seen which seemed to have foci of calcification (Figure 1). She underwent medical treatment with broad spectrum antibiotics. Four

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#### An unusual case of long-time intrauterine fetus retention

days following the onset of antibiotic therapy, cystoscopy and hysteroscopy were done. No abnormal finding was detected on cystoscopy. However, internal os of cervix was found to be obstructed on hysteroscopic examination, making access to uterine cavity impossible. Incredibly, some bony-like pieces were extracted by instrument from endocervical canal. According to this unexpected finding, total abdominal hysterectomy was planned for her. On surgery, pelvic cavity and pelvic organs were thoroughly examined for the presence of any probable fistula between uterus and its adjacent pelvic organs. Fortunately, no fistula was detected. The patient underwent total hysterectomy and bilateral salpingectomy. Uterus was opened coronally in operating room. Uterine cavity was occupied by lots of fetal bones. The estimated age of embryo was 23 weeks based on femur length (34 mm) (Figure 2). Surgical specimen was sent to pathology lab. On macroscopic examination, uterine cavity was encrusted by a thin plate of calcification covered by thick exudate. No intact endometrium was seen. Multiple fetal bones, including bones of extremities, ribs, and skull were buried within uterine cavity and completely isolated and walled off by calcified endometrial surface. Some long bones showed

superficial penetration into uterine mucosal surface (Figure 3). In serial sections of uterus, no grossly visible lesion was seen in myometrium. Uterine serosal surface seemed normal. Fallopian tubes were too thin, each having a diameter of 3 mm. On microscopic examination, endometrium was completely destroyed and replaced by granulation tissue with heavy lymphoplasmacytic and scattered eosinophilic infiltration. Surface of granulation tissue was covered by fibrinous exudate and neutrophilic debris and showed minor foci of squamatization. Sheets of calcification were seen within granulation tissue. In multiple sections prepared from endomyometrium, only very small residue of basalis endometrium was seen. Inner half of myometrium showed patchy infiltration of lymphocytes. Outer half of myometrium and uterine serosa were devoid of inflammatory cells. Endocervix and T-zone showed extensive erosion, granulation tissue formation, fibrinous exudate, and minor foci of calcification. Lamina propria of fallopian tubes mucosal folds showed patchy infiltration of lipofuscin and hemosiderin laden macrophages (pseudoxanthomatous salpingitis) (Figure 4).

Three days following hysterectomy, patient was discharged from hospital with good health condition.



Figure 1. Imaging findings (yellow arrow): A. Uterine ultrasound study revealing some linear calcified foci within uterine cavity; B. Plain abdominal X-ray showing some bones and air bubbles in pelvic region; C. Abdominopelvic computed tomography (CT) scan, showing a complex intrauterine mass with foci of calcification

#### M. Hajihashemi, et al.



Figure 2. Intrauterine retained bones of the dead fetus: A. Some bone fragments were extracted by instrument via endocervical canal during hysteroscopy; B. Uterus opened in operating room, showing retained bones of fetus (yellow arrow, femur; orange arrow, tibia and fibula; blue arrow, skull bones); C. Calculating fetal age using femur length. Femur length was 34 mm which is compatible with 23 weeks fetus



Figure 3. Macroscopic view of the uterus: encrustation of uterine cavity and multiple fetal bones are seen.



Figure 4. Microscopic sections: A & B. Sections from endometrium show complete destruction of endometrial tissue and its replacement by granulation tissue, fibrinous exudate, and sheets of calcification; C. Cervix also shows areas of erosion, granulation tissue formation, and fibrinous exudate, as well as minor foci of calcification; D. Pseudoxanthomatous salpingitis is evident as infiltration of lipofuscin and hemosiderin laden macrophages in lamina propria of tubal plica

## Discussion

Intrauterine retention of fetal bones, as a rare diagnosis, should be suspected when confusing complaints such as abdominal pain, irregular vaginal bleeding, dysmenorrhea, dyspareunia, and chronic pelvic pain are present. The most probable explanation for these symptoms is the inflammatory process induced by foreign body reaction in uterus (4). Since bones can act as an intra-uterine contraceptive device, infertility is the most common reported symptom. Therefore, despite its rarity, this condition should be considered as a cause of secondary infertility (5). Based on literature, these cases have been mainly discovered in low educated and low socioeconomic classes of societies and in areas with inadequate health care facilities (6). The phenomenon almost occurs when an intrauterine pregnancy has been neglected for a long period of time, resulting in absorption of whole structures of the fetus except bones over the time (6).

Transvaginal ultrasonography, abdominal CT scan, and hysterosalpingography are helpful in detecting bones in uterine cavity, but the best modality for evaluation of this condition is hysteroscopy. It allows definite diagnosis and simultaneous treatment (7). Removal of bones by hysteroscopy has been reported to lead to relief of symptoms and restoration of fertility in some cases (4,7).

The present case was receiving every-3-month-depomedroxy progesterone injection as birth control method. Therefore, we cannot consider infertility as one of the consequences of intrauterine retention of fetal bones in this patient. The most interesting aspect of this case report is complete ablation of endometrium, as confirmed by microscopic study, resulting from long standing intrauterine retention of fetal bones, which in turn had led to long standing inflammation and endometrial destruction. According to this finding, it seems that the patient had no chance of further successful pregnancies by reserving uterus. Since no fistula was detected between uterus and adjacent pelvic organs at surgery, air bubbles reported within uterine cavity on imaging studies could have been the result of infection induced by anaerobic bacteria.

Intrauterine retention of fetal bones, although rare, should be considered as a possibility in cases with unexplained abdominal pain, dysmenorrhea, dyspareunia, and long period infertility. Hysteroscopy is the best modality for diagnosis and may be enough for management of the condition in many cases. However, in complicated cases, hysterectomy would be inevitable.

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