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CASE REPORT

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Unusual intrauterine retained fetal skeletal bony fragments: A case report and review of the literature

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Abstract

An unusual finding in hysterectomy specimen. Associated with a history of abortion. Etiology remains unclear. Clinically it presents with infertility, chronic pelvic pain, menorrhagia, and vaginal discharge. Treatment is by hysterescopic excision.

KEYWORDS

fetal bony fragments, incomplete abortion, intrauterine, retention

1 | INTRODUCTION

Retention of fetal bones in the uterus is unusual complication of unsafe abortion. Patients may present with infertility, pelvic pain, menorrhagia, and vaginal discharge. We present a case of intrauterine retention of fetal bones in a mid-aged female. Abdominal hysterectomy was performed with a subsequent good outcome.

Retained intrauterine fetal bony fragments is an unusual and rare disease condition, often underdiagnosed,[1] with the reported incidence of 0.15% among diagnostic hysteroscopy.[2] The most common cause of retained intrauterine bony fragments is complication of unsafe abortion.[3] Other rare causes include osseous metaplasia, dystrophic calcification of the endometrium, and heteroplasia secondary to hypercalcemia, hyperphosphatemia, and hypervitaminosis.[4] Retained intrauterine bone tissue usually presents with gynecological problems such as infertility, menstrual irregularities, dysmenorrhea, vaginal discharge, and chronic pelvic pain. At times, patients may be incidentally diagnosed on a sonogram or hysteroscopy. The history of second trimester abortion is often denied particularly if it was done illegally and is usually obtained retrospectively after diagnosing retained intrauterine bony fragments.[3–5] Herein, we present a case of abnormal uterine bleeding secondary to retained fetal bone fragments.

2 | CASE PRESENTATION

A-45-year old female Para3, living 3, presented at our facility with chief complaint of abnormal vaginal discharge with foul smell and intermittent per vagina bleeding for 4 days. She also reported a history of amenorrhea for 1 year. Three days prior to the current hospital visit, she experienced lower abdominal pain that was associated with passing out bony tissue fragments per vagina. She attained menarche at the age of 16 years. She experienced irregular menstrual cycles that were lasting for 3 days. Her last normal menstrual period was over a year ago. Her past medical history revealed that she has been treated for epilepsy and HIV for the past 13 years on regular medication

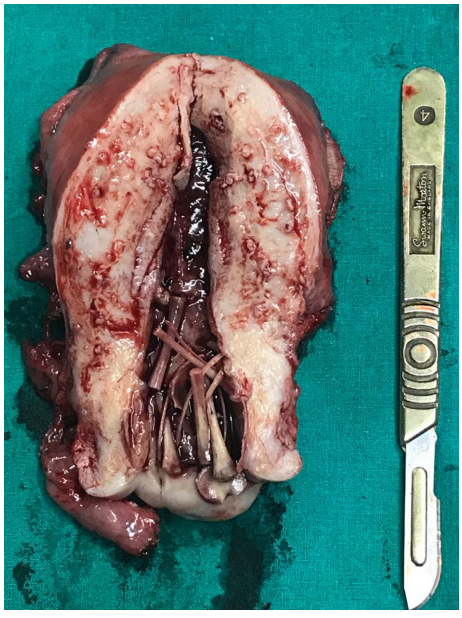
and has been treated for cervicitis recently. She denied history of abortion. Her deliveries were by lower segment cesarean section because of breech presentation, and the last delivery was 5 years ago.

On examination, she had essentially normal vital signs, a negative pregnancy test, and beta HCG test level of 3 mLU/mL. Abdominal pelvic USS revealed a relatively bigger uterine size with hyperechoic contents; there was no viable pregnancy appreciated. A rare differential diagnosis of uterine malignancy was entertained. Attempts to remove the bone fragments vaginally provoked intractable per vaginal bleeding that necessitated a prompt abdominal hysterectomy. Additionally, the patient was not planning for further fertility due to her age. The uterine specimen was submitted for histopathological analysis.

Gross examination of the specimen revealed about 16 fetal skeletal bony fragments filling the uterine cavity (Figures 1 and 2). Microscopically, the uterine specimen showed a hemorrhagic endometrium with bony remnants which were found to be mature, with degenerative and necrotic changes. The bony remnants showed mature degenerated osteocytes, bone tissue, and calcification (Figure 3). The patient was well postoperatively and was discharged home on day 4. She was apparently normal in subsequent follow-up visits.

3 | DISCUSSION

Encountering intrauterine fetal bony tissue fragments is an unexpected finding in a hysterectomy surgical specimen submitted to the department of pathology for histopathological examination. Case reports on intrauterine bone fragments retention have been reported.[6,7] In Nigeria, the incidence was 0.26% in 1002 hysteroscopies.[2] In 2018, Gainder et al., reported the incidence of 0.28% among women with infertility.[8] Basically, retained fetal bone is a rare complication of unsafe abortion. It is most common after evacuation of second trimester miscarriage.[9] It should be considered in all patients with infertility, abnormal uterine bleeding, dysmenorrhea, or vaginal discharge, dating from pregnancy termination.[8]

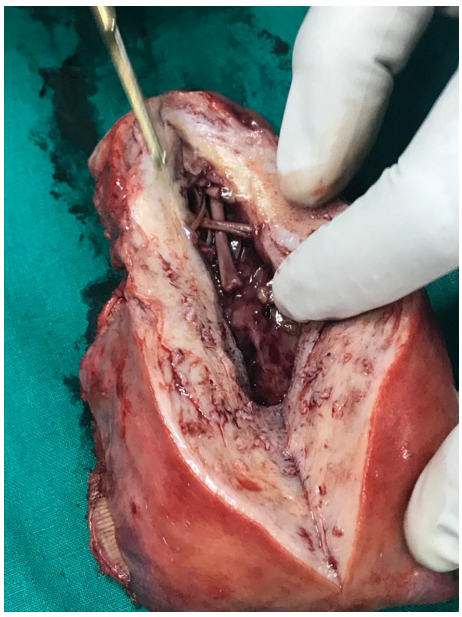


FIGURES 1 Gross views of the hysterectomy surgical specimen showing retained fetal bones in the endometrial cavity.

The most frequent etiologies of intrauterine fetal demise include genetic, infectious, thrombo-embolitic conditions as well as autoimmune diseases.[10] Similarly, environmental factors including tobacco and alcohol use, and endocrine diseases such diabetes have been implicated. The burden differs according to gestational age. Early miscarriage is basically result from genetic abnormalities such as aneuploidy and other chromosomal abnormalities, thrombophilic, and autoimmune. Second and third trimester miscarriages are essentially due to autoimmune, uterine anatomic abnormalities (polyps, leiomyomas, septae, cervical incompetence, etc.), maternal hypertension, thrombophilia, and infection/inflammation.[11] For our patient, it was difficult for us to determine the exact gestational age of the fetus. However, on the basis of the length and appearance of the bony fragments, we estimated that the fetus was likely to be 23 weeks or older.

Like it was the case in the index case, patients with retained uterine fetal bones usually present with nonspecific clinical presentations which may include abnormal vaginal bleeding, abnormal discharge, secondary infertility, chronic pelvic pain, and dysmenorrhea.[1,2] In the present case, abnormal vaginal bleeding was the chief complaint. A systematic review study involving 293 cases demonstrated that the mean ± SD age at presentation was 32.7 ± 8.9, and approximately 88% of patients had at least one prior surgical uterine evacuation related to pregnancy termination.[12] Although in the index case the history of abortion was denied, it is obvious that this was not valid. Perhaps the patient had experienced incomplete abortion without her knowledge. Alternatively, she might have decided not to disclose this fact.

Various pathogenic hypotheses have been proposed to explain the mechanisms that lead to bone tissue within the uterus. These include dystrophic ossification or osseous metaplasia of endometrium, heterotopic intrauterine bone, ossification of post-abortive endometritis, metastatic calcification, prolonged estrogen therapy after abortion, genital tuberculosis, and retained fetal bone are the commonly proposed theories.[13] Heterotopia implies the presence of mature tissue in an abnormal location. Bone



FIGURES 2 Gross views of the hysterectomy surgical specimen showing retained fetal bones in the endometrial cavity.

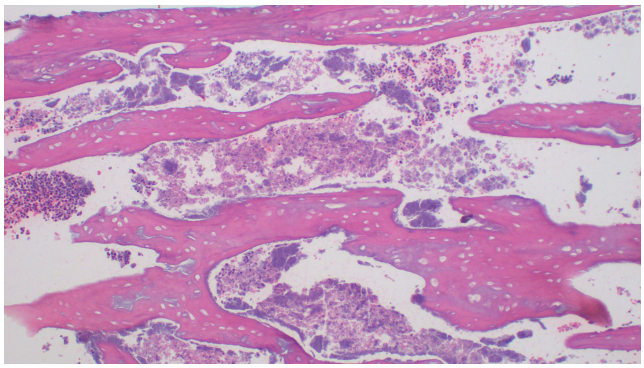


FIGURE 3 Histopathology of intrauterine bone tissue; demonstrating necrosis, and hypocellularity of the bone and marrow formation; hematoxylin and eosin staining 200 × original magnification.

is one of the most frequently cited heterotopic tissues. A history of antecedent induced abortion or curettage for spontaneous abortion is usually present. Retained fetal bone can cause multiple gynecological complaints such as metrorrhagia, menorrhagia, pelvic pain, foul-smelling leukorrhea, spontaneous elimination of bony fragments, and secondary infertility.[7,8] Unsafe abortion is one of the most neglected healthcare problems in developing countries. It is estimated that 97% of about 20 million unsafe abortions occur in these regions. High incidence is mainly due to restrictive abortion laws coupled with low level of awareness and use of contraception.[9]

The main suggested diagnostic modality for patients with this rare condition is ultrasonography.[4] In the present case, the sonographical findings were suggestive and later, the diagnosis was confirmed by gross and microscopic examination of hysterectomy specimen. Recommended treatment modality is hysteroscopic excision.[14] Nevertheless, non-hysteroscopic approaches such as biopsy, forceps, dilation, and curettage, and hysterectomy have been documented. In our patient, hysterectomy was performed partly because the patient had no desire of conceiving. Relief of symptoms following treatment is usually very rapid.

4 | CONCLUSION

Bone tissue within uterine cavity is a rare finding and it becomes an unusual cause of morbidity in females. Usually, it is associated with infertility and a past history of termination of pregnancy. Pathologists should be conversant of this rare entity. Women presenting with abnormal uterine bleeding following a pregnancy termination, retained fetal bones should be considered as one of the causes. Although rare, a high index of suspicion is the easier way to get correct diagnosis. Transvaginal ultrasound and radiological imaging of the pelvis can be used for diagnosis. Recommended treatment approach is removal of fetal bones. Patients become symptom free soon after treatment.

AUTHOR CONTRIBUTIONS

Alex Mremi: Conceptualization; data curation; investigation; resources; writing – original draft. **Yusuph Mwidibo:** Data curation; investigation; writing – review and editing. **Joseph Mlay:** Project administration; supervision; writing – review and editing. **Bariki Mchome:** Conceptualization; validation; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

All authors have declared that no competing interests exist.

DATA AVAILABILITY STATEMENT

None.

ETHICS STATEMENT

Ethical approval was waived by the authors' institution.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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