

## CASE REPORT

## Intrauterine Retention of Fetal Bones: An Unusual Cause of Infertility

Chukwuemeka O

**EZEAMA<sup>1,2</sup>**

Chukwuemeka C

**OKORO<sup>1</sup>**Joseph I **IKECHEBELU<sup>1,2</sup>**<sup>1</sup>Department of Obstetrics & Gynaecology

Nnamdi Azikiwe University

Teaching Hospital Nnewi

Anambra State, NIGERIA

<sup>2</sup>Department of Obstetrics & Gynaecology

College of Health Sciences

Nnamdi Azikiwe University

Nnewi, Anambra State

NIGERIA

**Author for Correspondence**Dr Chukwuemeka C **OKORO**

Department of Obstetrics &amp; Gynaecology

Nnamdi Azikiwe University

Teaching Hospital

PMB 5025 Nnewi

Anambra State, NIGERIA

Phone: +234 803 947 1821

E-mail: [emy4okoro@yahoo.com](mailto:emy4okoro@yahoo.com);[emy4okoro@gmail.com](mailto:emy4okoro@gmail.com)Received: September 1<sup>st</sup>, 2020Accepted: October 10<sup>th</sup>, 2020

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## INTRODUCTION

Infertility is the inability of a couple to achieve conception after one year of unprotected intercourse.<sup>1,2</sup> It is a major cause of marital disharmony because of the high premium placed on child bearing especially in developing countries. Some societies like ours consider child bearing as the major

reason for marriage, and without children no meaningful social or economic progress is considered worthwhile.<sup>3</sup> In developing countries, most of the cases of infertility are secondary infertility attributable to complications of unsafe abortion, sexually transmitted infections and puerperal pelvic infections. The common cause of infertility

**ABSTRACT**

Couples presenting with infertility face peculiar sociocultural problems, these are especially evident in African countries where a very high premium is placed on children and childbearing.

We report a case of a 30-year-old nullipara with a 3-year history of secondary infertility who on investigation was found to have fetal bones from an earlier incomplete abortion. She achieved pregnancy within three months of endoscopic removal of the fetal bones.

in patients with previous termination of pregnancy is tubal disease resulting from tubal infection, uterine synechia from excessive curettage (Asherman syndrome) or endometritis.<sup>4</sup> Rarely, retained foetal bones may be the culprit in this group of patients.<sup>5</sup>

#### CASE SUMMARY

The patient is a 30-year-old P<sub>0</sub><sup>+1</sup> woman with a 3-year history of secondary infertility. She had vaginal instrumentation for termination of pregnancy as an adolescent girl 10 years earlier at gestational age of 14 weeks by a quack. This resulted in an incomplete abortion with bleeding per vagina. She had a repeat uterine evacuation by the same person following which the bleeding stopped and she became apparently well.

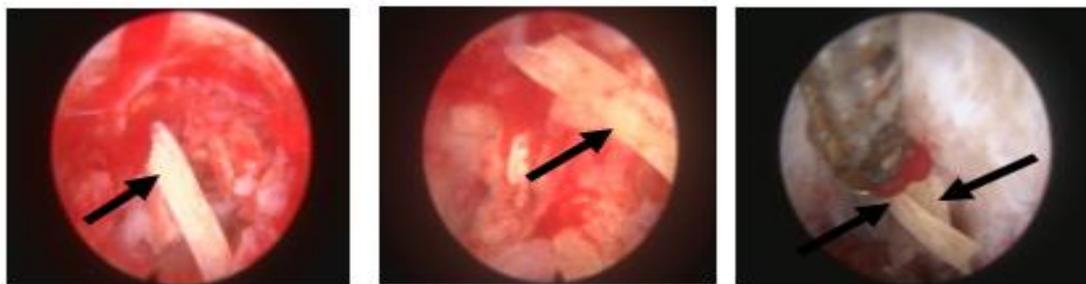
The couple had routine evaluation for infertility and findings were normal except for abdominopelvic ultrasound scan which showed a normal sized uterus, hyper-reflective endometrium with two areas of calcification casting posterior acoustic shadows; one measured 16x7mm while the other was 10x8mm. An impression of

calcified intracavitary object suspected to be calcified uterine fibroid was made by the sonologist.

The patient was counselled and she had hysteroscopic assessment which revealed three pieces of foreign body lying within the upper and mid portions of the endometrial cavity. Hysteroscopic forceps was used to extract the hard tissues within the endometrial cavity. They measured 16x7mm, 16x5mm, 10x8mm and were sent for histopathological study which identified them as fetal bones. Macroscopy of the specimen showed decalcified tiny chips of grayish white substance while microscopy showed woven bone with marrow spaces containing red blood cells and neutrophil polymorphs.

After the removal of the foreign bodies, she achieved pregnancy following her second menstrual flow (within 2 months) without further intervention. She had emergency caesarean section at gestational age of 36 weeks on account of severe preeclampsia in a twin gestation with good outcome.

**Figure 1.** Hysteroscopic views of the retained fetal bones. The first 2 plates show various views of the fetal bones lying in the endometrial cavity. The last plate shows the hysteroscopic forceps being used to extract the bones.



#### DISCUSSION

Intra-uterine foreign bodies have been identified as rare causes of infertility, as is illustrated in our case report.<sup>6</sup> Such foreign bodies are especially seen in women who have had a termination of pregnancy. This case report illustrates one of the causes of

infertility especially in women who have had a poorly managed termination of pregnancy. Examples of foreign bodies which have been reported to cause infertility include fetal bones, non-absorbable suture materials (following caesarean section, cervical cerclage or myomectomy),

intrauterine contraceptive device (complete or part of it), broken laminaria tents, tips of curettes and other instruments.<sup>6,7</sup> Endometrial osseous metaplasia has also been described as a cause of infertility.<sup>8</sup> Dystrophic calcification, heterotopia, ossification of post-abortion endometritis, metastatic calcification, and prolonged oestrogen supplement after abortion have all been cited as explanations for the endometrial osseous metaplasia.<sup>8</sup>

Retained fetal bone is a rare complication that can follow abortion especially when performed in the late first or early second trimester as seen in our patient. This is in addition to the very high risk of excessive haemorrhage and uterine perforation following the evacuation of the uterus at advanced gestations. Retention of fetal bones is more likely to complicate termination of late pregnancy when such termination is done by dilatation and curettage.<sup>9</sup> Retention of fetal bones in the uterus could also result following the removal of a macerated fetus or destructive deliveries of a dead fetus. The fetal bones can be retained freely in the endometrial cavity or embedded, totally or partially, in the myometrium.<sup>10</sup> A large numbers of women with intrauterine fetal bones have infertility. However, its overall contribution to infertility is very meagre.<sup>7,11</sup>

Patients with retained foetal bones may present with menstrual bleeding abnormalities (like amenorrhoea, irregular bleeding, intermenstrual bleeding and menorrhagia), abnormal vaginal discharge, dysmenorrhoea, chronic pelvic pain, passage of bony fragments vaginally and secondary infertility.<sup>2,4,6,7,9</sup> Sometimes, retained bones are completely asymptomatic, only to be discovered following pelvic ultrasound scan as part of routine gynaecological assessment.<sup>12</sup> Our patient presented with secondary infertility.

The risk of infertility depends on the location of the retained piece of bone. Completely intramural bony fragments do not seem to compromise fertility; conversely, retained fetal bones lying freely in the endometrial cavity or those that are only partially embedded are associated with a high risk of infertility.<sup>12</sup> Retained bones within the endometrial cavity cause an increase in the menstrual fluid prostaglandin E and prostacyclin which can cause vasodilatation and myometrial relaxation. The pattern is similar to that seen in intra-uterine contraceptive device (IUCD) users resulting in local release of prostaglandins and poor endometrial implantation especially when it is in the fundal region of the uterus.<sup>13,14</sup> Another speculated mechanism through which it causes infertility is by induction of uterine synechia and hence impairment of implantation.<sup>9</sup>

In addition to detailed history and physical examination, useful investigations in the management of patients with intrauterine foreign body include pelvic ultrasound scan (preferably transvaginal scan), saline infusion sonography, computed tomography (CT) scan, endometrial curettage and hysteroscopy.<sup>9,12</sup> Hysterosalpingography (HSG), which is routine in infertility evaluation for outlining the endometrial cavity and in determining the state of the fallopian tubes, is of limited usefulness here as the opacity of the contrast will blend with that of the retained fetal bones. Elford and Claman reported that hysteroscopy can miss the diagnosis even when it is visible on ultrasound scan.<sup>15</sup> This is especially true when the fetal bone is completely embedded in the myometrium. Other components of the infertility work up should be carried out since other causes of infertility could co-exist with the foreign body.

Whenever an intra-uterine foreign body is suspected, it is good practice to do a hysteroscopy to confirm the diagnosis and possibly retrieve the foreign body as was done in our case. Hysteroscopy is the standard modality for management of this entity, both for confirmation and for operative removal.<sup>9,16</sup> In places where hysteroscopy is not available, or in cases where the foreign body is not visualised at hysteroscopy, ultrasound guided dilatation and curettage can be used to evacuate the uterus.<sup>15</sup> Where portable ultrasound machine is not available, blind dilatation and evacuation of the uterus and postoperative ultrasound scan to confirm complete removal of the bony pieces, is fairly satisfactory.<sup>9</sup> Obviously, ultrasound guidance can only be employed if the bony pieces were visible by ultrasound ab initio.

As was seen in our case, reproductive outcome is good following removal. The presence of fetal bones in the uterine cavity act in a similar manner to an IUCD, therefore, removal of the foreign body should be followed by spontaneous pregnancy if there are no other contributory factors to the infertility. In their series, Moon *et al.* described 10 spontaneous pregnancies following intrauterine bone removal in 11 patients.<sup>13</sup> The only patient in their series who was not pregnant had tubal damage.

Graham *et al.* noted that within a 7 year period, 11 out of the women presenting to King's College Hospital Assisted Conception Unit have intrauterine foreign body.<sup>17</sup> All the cases were West African women. All the women had a history of one or more terminations at gestations between 10 and 26 weeks in their countries of origin. A possible explanation for this condition being more common in West Africans is the terminations of pregnancies by inexperienced practitioners, unqualified persons and in

unhygienic settings. In addition, the procedure is more likely to be done later in pregnancy with greater risk of being incomplete. In their series, following the removal of foreign body, eight women conceived spontaneously resulting in 12 pregnancies: one ectopic, two miscarriages, and nine deliveries. One patient was lost to follow up. Medical practitioners should be aware of the complication of mid-trimester dilatation and evacuation and avoid it especially now that safe, effective and cheap prostaglandins are available. Even when dilatation and evacuation is used under certain conditions, transvaginal pelvic ultrasound scan should be done to confirm complete evacuation. Practitioners especially those treating West African women with infertility should be aware of this condition and include transvaginal ultrasound scan and hysteroscopy in their investigations. Again, this is another reason for recommending hysteroscopy to be included in the work up of patients with infertility as it could reveal uncommon causes of infertility.

Though the retention of fetal bones within the uterine cavity is rare, it is a treatable cause of secondary infertility. The diagnosis should be considered if highly echogenic material is identified on transvaginal ultrasound scan regardless of any history of previous pregnancy, pregnancy termination or the interval between the preceding termination of pregnancy and presentation. Confirmation and removal is usually accomplished using hysteroscopy. A high live birth rate may be achieved following removal of the retained bone.

#### **Declaration of Patient Consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient

understood that her names and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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