ORIGINAL ARTICLE

TITLE: RETAINED FOETAL BONE FRAGMENT MASQUERADING AS FOREIGN BODY CAUSING SECONDARY INFERTILITY

ABSTRACT

BACKGROUND - Termination of pregnancy is on the rise in India, due to variety of safer abortion techniques, and readily available facilities. When conducted correctly, pregnancy termination (abortion) is a relatively safe treatment. Pregnancy termination is linked to immediate and delayed problems such as haemorrhage and uterine perforation, as well as Asherman's syndrome, menstrual irregularities, and secondary infertility. However, difficulties can occur even when procedures are carried out with the utmost care. The retention of foetal products after a mid-trimester abortion is an uncommon abortion complication that can lead to subsequent infertility.

In rare situations, retained foetal bone fragments can induce acute pelvic inflammatory disease, regardless of how long it has been after the abortion. Recurrent vaginitis or endometritis that is resistant to empirical antibiotic treatment can also be symptoms of this illness. In such circumstances, the possibility of a foreign body in the uterine cavity should be considered. Ultrasonography should be used to evaluate such individuals first, as it is crucial in the differential diagnosis of these situations.

CASE –
We hereby present a case of a 25-year-old lady who had an abortion at the 14th week of pregnancy due to anencephaly in the baby and thereafter was unable to conceive for the next five years. The presence of a suspected foreign body or endometrial calcification in the upper uterine cavity was discovered during a transvaginal Ultrasonographic examination and was removed hysteroscopically. Following the hysteroscopic removal of bone piece, the patient was able to conceive. Remaining bone pieces from a prior mid-trimester abortion thus should be added to the list of secondary infertility differentials.

OBJECTIVES –
We evaluated the relevant causative factors, radiographic abnormalities, treatment strategies and clinical recovery in secondary infertility

CONCLUSIONS –
Secondary infertility caused by retained foetal bones is an uncommon occurrence that an obstetrician should be aware of while evaluating patients who have had a previous mid-trimester abortion. For initial diagnosis, transvaginal ultrasonography is a useful tool. Hysteroscopy is useful for both diagnostic and therapeutic purposes. The majority of these patients should be able to conceive again. In the post-abortion phase, all patients who had a second-trimester abortion should get a trans-vaginal ultrasound to confirm full evacuation of all the products of conception.

KEYWORDS –
Foreign body, secondary infertility, ultrasonography, hysteroscopy, bony fragment

INTRODUCTION –

The presence of intrauterine bone structures after an abortion is a rare case. The precise occurrence is uncertain. In developing nations, illegal abortion is one of the most under-reported health concerns. Retained products of pregnancy are a relatively uncommon complication of unsafe abortion. In patients undergoing diagnostic hysteroscopy, the prevalence is around 15 percent(Foreste et al., 2021). There are no obvious signs or symptoms in some patients. Subfertility, abnormal vaginal discharge,irregular uterine bleeding, dyspareunia,lower abdominal pain, dysmenorrhoea, and spontaneous passage of bony fragments have all been studied extensively.

Advances in medicine have made it safer to conceive and give birth, both in terms of diagnosis and care. The right to terminate or abort a pregnancy owing to foetal anomalies spares the unborn child and the family the pain that would otherwise occur if the child were born. Even though terminating a pregnancy is a relatively safe treatment, it can result in immediate consequences including haemorrhage, uterine perforation, and cervical trauma, as well as late complications such infections, bleeding, menstrual abnormalities, and uterine synechiae (Sajadi-Ernazarova & Martinez, 2021). There is a danger of subsequent infertility as a result of a complicated abortion because 50 percent of patients seeking surgical termination are young adults.(Atrash & Hogue, 1990).

If the abortion is performed in the second trimester or the foetus is removed by destructive techniques, a few pieces of the foetus may unintentionally be left behind in the uterine cavity. Pelvic pain, irregular bleeding, infection, dyspareunia, and even the passage of foetal components per vaginum are all possibilities in this scenario. Women may experience secondary infertility as a result of retaining foetal components. Retained foetal parts are most likely to induce uterine response, such as vaginal discharge and/or discomfort, as well as subsequent infertility in the majority of these patients.(Srofenyoh et al., 2006).

A case of secondary infertility due to foetal bone pieces lingering in the uterine cavity is presented. There may be no symptoms or may have symptoms such as dysmenorrhoea, vaginal discharge, or irregular uterine bleeding in the patient. Using hysteroscopic procedures, the identification and treatment of such a situation can be achieved successfully.

CASE PRESENTATION -

For infertility screening and treatment, a 25-year-old lady was referred to the AVBRH Hospital's Gynaecology out-patient clinic. Her medical history revealed that she was suffering from secondary infertility.
She had been suffering from dyspareunia for two months. She had a history of second trimester abortion. On an ultrasonographic test two years ago, the foetus was discovered to have anencephaly. The pregnancy was terminated, and labour was induced. She saw a number of doctors and had a variety of procedures over the course of the last five years, but she was unable to conceive. Our hospital was then referred to her.
Both couples were exposed to infertility investigations. The man's sperm count was normal. There were no abnormalities in the woman's hormonal profile.

The patient appeared to be of average build on physical inspection. Her vital signs were all within normal limits. Pelvic examinations were performed vaginally and abdominally and found to be unremarkable in both cases. Her Transvaginal sonography showed the presence of linear calcific density hyperechoic area measuring approx 12 mm noted embedded within the upper endometrium, horizontally oriented , it showed a twinkling artefact on color Doppler imaging which was suspected to be foreign body or endometrial calcification[Figure 1]. The findings were validated by our hospital's consulting radiologist. A diagnostic hysterolaparoscopy with foreign body removal was scheduled.

The procedure was performed under general anaesthesia that revealed the existence of a cylindrical structure in the uterus that appeared like a miniature long bone fragment under vision[Figure 2,3,4]. The fragments were removed under vision using dilation and curettage. The bony piece fragment was retrieved from the patient’s uterus.. The patient was kept on intravenous antibiotics for 24 hours following the hysterolaparoscopy procedure. The removed material was sent to a pathologist for histological analysis, and the pathologist identified the material as bone. On gross pathological examination revealed a single, irregular, whitish thread like tissue piece measuring 0.5 x 0.5 cm[Figure 5]. Section from given specimen shows histopathological features suggestive of bone trabeculae of normal morphology probably retained products of conception[Figure 6]. Her first check-up took place after she had her first period. Following that, monthly follow-ups were carried out. Her menstrual cycle was unaffected by the operation, and 5 months after the hysteroscopic removal of bone fragments, she was able to conceive. As a result, the preserved foetal bony fragments confirmed our suspicions of secondary infertility.

Figure 1 – TVS showing linear calcific deposit with endometrium

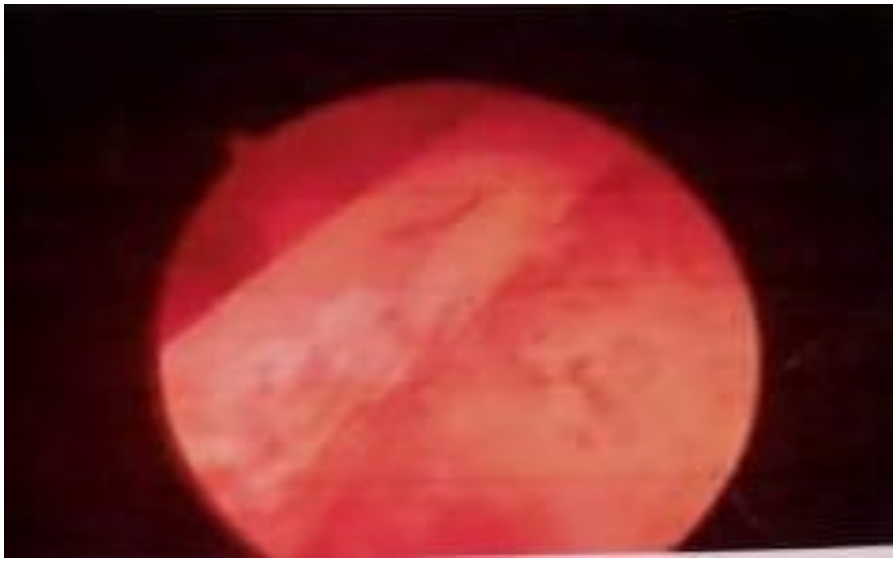

Figure 2 – hysteroscopic view of cylindrical structure filling the cervical canal and endometrial cavity

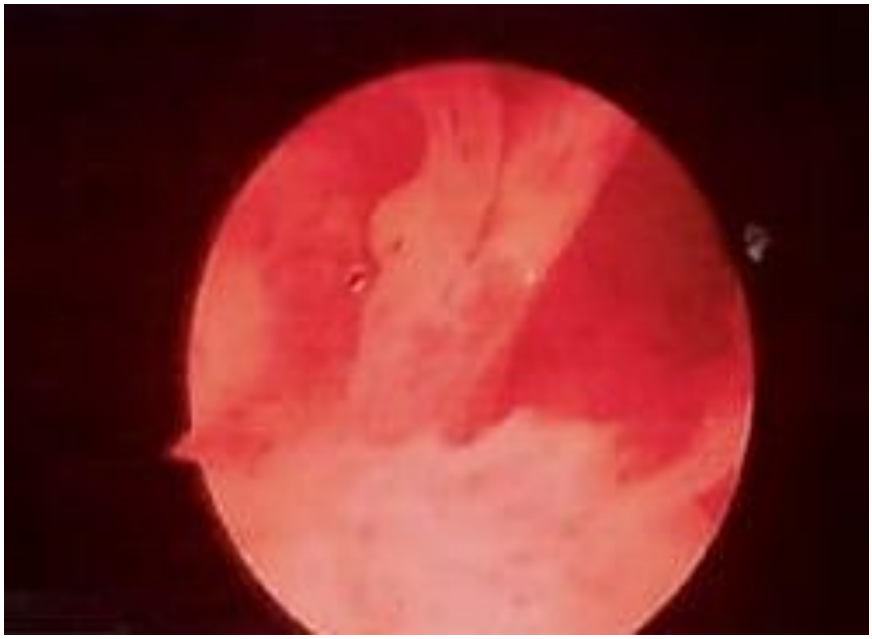

Figure 3 : coral-like thin calcified bony material on hysterescopic evaluation

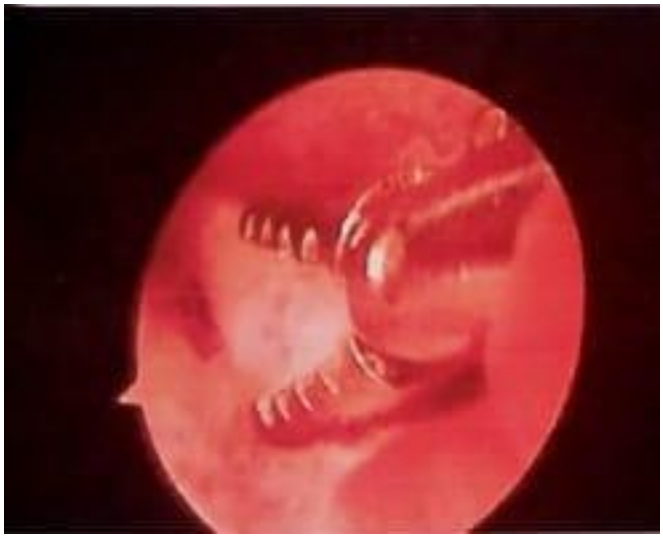

Figure 4 : hysteroscopic removal of foreign material

Figure 5 : Gross view of hysteroscopically retrieved intrauterine fetal bones

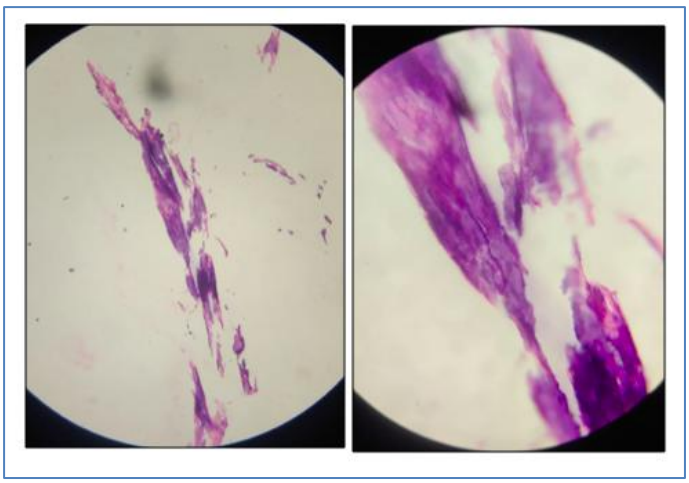

Figure 6. Histopathologic image showing bony trabeculae

DISCUSSION –

Endometrial ossification has been reported in many studies. It's assumed to be generated primarily by foetal bone fragments, but it can also be caused in some cases by metaplasia of mature endometrial stromal cells as a result of persistent inflammation or trauma.

According to the Roth and Taylor hypothesis ,chronic inflammation or trauma can cause mature endometrial stromal cells to undergo metaplasia into bone tissue. According to the researchers, the bone fragments cause infertility by acting as an intrauterine device, causing an increase in endometrial prostaglandins and blocking the implantation process.

"The bone remains had been working as an intrauterine contraceptive device and that a pregnancy following their removal is more than coincidence," Naftolin concluded in a related case of infertility (Bakhshi et al., 2004). Bone pieces in the uterine cavity could cause myometrial contractions, which would further embed the fragments in the myometrium. As a result, spontaneous evacuation becomes less frequent over time, allowing normal endometrium to overgrow the firmly buried fragments.

In addition to their infertility, many women with retained foetal bone will experience symptoms such as menometrorrhoea, dysmenorrhoea, vaginal discharge, pelvic pain, and spontaneous elimination of bony fragments in the menses (Chervenak et al., 1982). Lewis et al. evaluated menstrual blood volume and prostaglandin E2 concentrations in a woman with infertility and menorrhagia before and after the extraction of retained foetal bones. After the retained bone was removed, he discovered that the menstrual volume and total prostaglandin content reduced by 50%(Lewis et al., 1990).

An unusual complication of induced abortion, spontaneous intrauterine foetal death, and missed abortion is prolonged intrauterine retention of foetal bone sections. Although abortion is a safe procedure when done correctly, it does come with its own set of risks. Incomplete uterine evacuation can lead to a variety of problems, including the uncommon complication of secondary infertility, as we've seen in this case. In order to clarify the potential cause of infertility in such situations, variety of theories has been suggested in the literature. According to the theory, retained foetal tissues from a previous incomplete abortion can cause dystrophic calcification and ossification of the foetal tissue, which leads to bone formation. Chronic inflammation and tissue death may also promote heterotopic bone development in the mature uterine stroma (Mishra et al., n.d.). In the circumstance at hand, both options are quite unlikely. The extraction of a reasonably big tubular bone that looked like a foetal long bone indicated that it came from a foetus and not from dystrophic or heterotrophic

calcification. The diagnosis was also verified by histopathologic study of the retrieved fragments.

Retained foetal bone has been proposed as an intrauterine contraception device that alters the uterine environment and prevents implantation. Other theories for why retained foetal bones induce infertility include obliteration of the uterine cavity, which prevents implantation, and toxicity of bone particles on an implanted embryo(Singla et al., 2012).

Obtaining a detailed history is essential when examining a patient with secondary infertility, regardless of the aetiology. In all cases, TVS should be performed to assess the completeness of evacuation, and hysteroscopy should be scheduled if any worrisome echoes are detected.

In a patient with retained foetal bones, hysteroscopic excision of the bony parts followed by curettage has been shown to be an adequate treatment for infertility. The menses returned after the bone piece was removed in this case, and the patient conceived in six months.

For successful treatment of secondary infertility, it is critical to recognise foreign bodies in the uterine canal, such as a piece of intrauterine contraceptive (after removal failure), suture after caesarean section, or cylindrical and flat tiny bones after abortions. During the years 1992–1993, Kazakov et al. reported removing 69 foreign entities with a wire loop resectoscope (Kazakov et al., 1994). Sixty-one of the foreign bodies were devices that were implanted in the uterus. The remaining eight women (11.9%) had their foetal bones extracted. TCRF (transcervical resection of foreign bodies) is a safe and effective procedure. For a successful TCRF, there must be enough cervical canal distension, as well as the right equipment and methodology. B-mode ultrasonography is the first choice for monitoring TCRF because it is non-invasive and effective.
We believe that the current report adds to the pool of knowledge of practising obstetricians and makes them aware of the importance of examining retained bones from past abortions when addressing secondary infertility differentials.

Pregnancy terminations are becoming more common for a variety of reasons. Despite the fact that the safety quotient has increased with time, issues still occur. They may stay asymptomatic, as in our case, making diagnosis challenging. It works as an intrauterine contraceptive device and prevents conception, as evidenced by the fact that the lady pregnant four months after the bone pieces were removed. Secondary infertility should be treated with scepticism, and a hysteroscopy should be conducted in the same session for precise diagnosis and treatment. In the literature, the presence of intrauterine bony structures has been described. Retained foetal bone fragments are the most prevalent cause of these structures, but some cases may be caused by metaplasia of mature endometrial stromal cells in response to persistent inflammation or trauma.

It's worth noting that in cases of uterine malformations, the presence of retained foetal bones may be more common (Chervenak et al., 1982; Melius et al., 1991). Before instrumentation, attention must be made to understand the anomaly in these

circumstances. This will reduce the danger of uterine perforation and make thorough removal of the fragments easier.

Rather to the traditional laparohysteroscopy, several studies now emphasise the role of transvaginal sonographic diagnosis of the female genital tract as a first-line diagnostic technique. During the couple's work-up, a basic ultrasound-based strategy to investigating the infertile patient can be used effectively as an initial examination modality. A normal endometrium is indicated by a regular myometrial-endometrial interface and a homogeneous endometrial structure on TVS that corresponds to the menstrual cycle phase and eliminates the need for diagnostic hysteroscopy. TVS can be used to screen patients for hysteroscopy as a first step in the diagnostic process.

CONCLUSIONS -

All patients with infertility, dysfunctional uterine bleeding, dysmenorrhea, or other signs related to a pregnancy or pregnancy termination should be evaluated for retained foetal bones(Gainder et al., 2018; Wong et al., 2002). TVS should be performed in all women presenting as secondary infertility who had a prior pregnancy that progressed past 11 weeks gestation and resulted in spontaneous or surgical abortion should have a transvaginal sonogram done. Prolonged retention of intrauterine bone is a recognized cause of secondary infertility. TVS can also be used to track patients who have had their bony fragments surgically removed. Few bony fragments may be lodged in the endometrium or myometrium, making curettage difficult to identify. Because of the accessibility of care and the favourable postoperative prognosis, all women with secondary infertility who have had an abortion should have a thorough transvaginal sonographic review of the endometrium. This examination would not only expose irregular endometrial pathology, but it may also reveal uterine abnormalities that were previously undetected. In both determining the diagnosis and extracting bony fragments, a diagnostic hysteroscopy accompanied by an operative procedure using a wire loop resectoscope is invaluable. The reintroduction of the hysteroscope to record that the cavity is clear after attempts at bone removal are complete is an integral part of the procedure. Following the removal of the bony fragments, it is expected that fertility will be regained and symptoms will improve.

Disclaimer regarding Consent and Ethical Approval:

As per university standard guideline, participant consent and ethical approval have been collected and preserved by the authors

CONFLICT OF INTEREST –

The authors stated no conflict of interest.

REFERENCES-

Atrash, H. K., & Hogue, C. J. (1990). The effect of pregnancy termination on future reproduction. *Bailliere's Clinical Obstetrics and Gynaecology*, *4*(2), 391–405. https://doi.org/10.1016/s0950-3552(05)80234-2

Bakhshi, P. S., Allahbadia, G. N., Kaur, K., & Virk, S. P. S. (2004). Hysteroscopic removal of intrauterine retained fetal bones. *Gynecological Surgery*, *1*(3), 159–166. https://doi.org/10.1007/s10397-004-0033-0

Chervenak, F. A., Amin, H. K., & Neuwirth, R. S. (1982). Symptomatic intrauterine retention of fetal bones. *Obstetrics and Gynecology*, *59*(6 Suppl), 58S-61S.

Foreste, V., Gallo, A., Manzi, A., Riccardi, C., Carugno, J., & Sardo, A. D. S. (2021). Hysteroscopy and Retained Products of Conception: An Update. *Gynecology and Minimally Invasive Therapy*, *10*(4), 203. https://doi.org/10.4103/GMIT.GMIT_125_20

Gainder, S., Arora, P., & Dhaliwal, L. (2018). Retained Intrauterine Bony Fragments as a Cause of Secondary Infertility in a Tertiary Level Indian Hospital. *Journal of Human Reproductive Sciences*, *11*, 286. https://doi.org/10.4103/jhrs.JHRS_33_18

Kazakov, null, Khankoev, null, & Pererva, null. (1994). Results of Hysteroscopic Method of Foreign Body Removal out of Uterus Cavity. *The Journal of the American Association of Gynecologic Laparoscopists*, *1*(4, Part 2), S16. https://doi.org/10.1016/s1074-3804(05)80918-1

Lewis, V., Khan-Dawood, F., King, M., Beckmann, C., & Dawood, M. Y. (1990). Retention of intrauterine fetal bone increases menstrual prostaglandins. *Obstetrics and Gynecology*, *75*(3 Pt 2), 561–563.

Melius, F. A., Julian, T. M., & Nagel, T. C. (1991). Prolonged retention of intrauterine bones. *Obstetrics and Gynecology*, *78*(5 Pt 2), 919–921.

Mishra, N., Bharti, R., Mittal, P., Suri, J., & Pandey, D. (n.d.). Retained Intra-uterine Foetal Bones Resulting in Secondary Infertility: A Case Report. *Cureus*, *10*(5), e2575. https://doi.org/10.7759/cureus.2575

Sajadi-Ernazarova, K. R., & Martinez, C. L. (2021). Abortion Complications. In *StatPearls [Internet]*. StatPearls Publishing. https://www.ncbi.nlm.nih.gov/books/NBK430793/

Singla, A., Gupta, B., & Guleria, K. (2012). First Trimester Abortion: A Rare Cause of Intrauterine Bony Spicules. *Case Reports in Obstetrics and Gynecology*, *2012*, 701021. https://doi.org/10.1155/2012/701021

Srofenyoh, E., Addison, M., Dortey, B., & Kuffour, P. (2006). Intrauterine Retained Fetal Bones as a Cause of Secondary Infertility. *Ghana Medical Journal*, *40*(3), 105–109.

The potential consequence of early recognition and treatment of retained fetal bony fragments: Journal of Obstetrics and Gynaecology: Vol 22, No 4. (n.d.). Retrieved January 19, 2022, from https://www.tandfonline.com/doi/abs/10.1080/014436102320261177

Wong, S. F., Lam, M. H., & Ho, L. C. (2002). Transvaginal sonography in the detection of retained products of conception after first-trimester spontaneous abortion. *Journal of Clinical Ultrasound: JCU*, *30*(7), 428–432. https://doi.org/10.1002/jcu.10086