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REVIEW ARTICLE

P. S. Bakhshi · Gautam N. Allahbadia · Kulvinder Kaur · S. P. S. Virk

Hysteroscopic removal of intrauterine retained fetal bones

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Abstract Fetus papyraceus is a well-recognized complication of missed abortion or intrauterine fetal death. However, prolonged intrauterine retention of parts of the fetal skeleton is not adequately reported in the literature. Prolonged retention of intrauterine bone is a recognized cause of secondary infertility. Literature regarding endometrial ossification contains more than 80 cases, with 80% occurring after pregnancy. Common symptoms include menorrhagia, menometrorraghia, pelvic pain and infertility. Several theories have been proposed to explain osseous tissue within the uterine cavity. The most widely accepted is that ossification represents retained fetal bones following spontaneous, missed, incomplete or therapeutic abortion, suggesting that the fetus achieved at least 12 weeks gestation in order for endochondral ossification to occur. In contemporary gynecology, transvaginal sonographic diagnosis of retained fetal bones is made and the bones removed with the help of a resectoscope. Hysteroscopy is invaluable in achieving a cure for these patients without resorting to more aggressive surgery. Gonadotropin-releasing hormone analogues (GnRHa) have many new uses in gynecology. Several clinical studies have demonstrated a mean decrease in uterine volume after about 3–6 months of therapy. These patients might be candidates for GnRHa therapy so that a smaller corpus with an atrophic endometrium might aid in hysteroscopic retrieval of the retained bony fragments.

P. S. Bakhshi · K. Kaur · S. P. S. Virk
Rotunda-Virk Center For Human Reproduction,
Jalandhar, India

G. N. Allahbadia (✉)
Rotunda-The Center For Human Reproduction,
Mumbai and Rotunda-Virk Center for Human Reproduction,
Jalandhar, India
e-mail: drallah@vsnl.com
Fax: +91-22-26553000

G. N. Allahbadia
672 Kalpak-Gulistan, Perry Cross Road, Near Otters Club,
Bandra (West), Mumbai 400050, India

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Introduction

Retained intrauterine fetal bones may cause infertility. Dawood and Jarrett [1] postulated that retained fetal bones may act as an intrauterine synechia or intrauterine device, increasing endometrial prostaglandin F2alpha and preventing implantation, resulting in infertility. Also, the reactive endometritis caused by the bones probably interferes with blastocyst implantation. The diagnosis was traditionally suspected from filling defects on hysterosalpingography, with the definitive diagnosis made on blind curettings. Therapy had generally been restricted to curettage or hysterectomy [2, 3, 4]. Allahbadia et al. first published a paper where the diagnosis was made by transvaginal sonography, which helped in the exact localization of retained fetal bone as to its location in the uterus and ruling out deep invasion in the endomyometrium [5]. We propose that if on transvaginal sonography we detect a hyperechogenic shadow in infertility patients, especially those who also have irregular bleeding or symptoms dating from a pregnancy or pregnancy termination, we should think in terms of retained fetal bones. We can stretch the spectrum of transvaginal sonographic diagnosis further by doing a saline infusion sonohysterography (SIS) in all such cases where 1–5 ml saline is injected through a 5F Hysterosonography Elliptosphere Catheter set (Ackrad Laboratories, Inc, USA) into the uterus to distend the endometrial cavity [6]. This technique allows detailed visualization of the uterine cavity in both longitudinal and transverse planes, and the endometrium can be evaluated for the presence of polyps, submucous fibroids, intrauterine synechiae and foreign bodies [6]. Also, the use of a resectoscope in these cases as opposed to operative hysteroscopic sheaths [7] and alligator forceps [8] brings about ease of surgical

removal of the partially or completely embedded bony spicules and cartilage fragments.

Etiopathology

Roth and Taylor [9] hypothesized that without a previous pregnancy loss, metaplasia of mature endometrial stromal cells into bony tissue occurs in response to chronic inflammation or trauma. Sporadic cases without antecedent pregnancy support this theory [9, 10, 11, 12]. Over the past few decades there have been several case reports describing endometrial ossification [13, 14, 15, 16, 17]. It is most commonly believed to be due to retained fetal bone fragments, but some cases may be due to metaplasia of mature endometrial stromal cells in response to chronic inflammation or trauma [9]. Recently, it has been suggested that the incidence of this complication after induced or spontaneous abortions is underestimated in the literature [10]. In the early 1990s, Melius et al. [7] found more than 50 cases in the literature, with 80% of them occurring after pregnancy.

Most cases of endometrial ossification are discovered because of prolonged secondary infertility after a late therapeutic abortion. Researchers suggested that the bone fragments cause infertility by acting like an intrauterine device, increasing endometrial prostaglandins and preventing implantation [1]. Commenting on a similar case of infertility, Naftolin concluded in 1999 that "the bone remnants had been acting like an intrauterine contraceptive device and that a pregnancy following their removal is more than coincidence" [18].

We speculate that bone fragments held within the uterine cavity might stimulate myometrial contractions, which may further embed the fragments deep into the myometrium. Thus, spontaneous expulsion becomes less likely with passing time and normal endometrium can then overgrow the deeply embedded fragments.

Many women with retained fetal bone will have symptoms of menometrorraghia, dysmenorrhea, vaginal discharge, pelvic pain and spontaneous elimination of bony fragments in the menses in addition to their infertility [8]. Lewis et al. measured menstrual blood volume and prostaglandin E2 concentrations before and after the removal of retained fetal bones in a woman with infertility and menorrhagia. He found the menstrual volume and total prostaglandin concentration decreased by 50% after the retained bone was removed [3].

It is interesting to note that the presence of retained fetal bones may be more common in cases of uterine anomalies [7, 8]. In these cases, care must be taken to understand the anomaly before instrumentation. This will minimize the risk of uterine perforation and will help to facilitate complete removal of the fragments. Wetzels et al. [10] described a case of endometrial ossification in a septate uterus that was extremely difficult to evacuate and required a number of serial procedures.

Fig. 1 Hysteroscopic view of retained bone fragments, most probably fragments from a long bone

Diagnosis

Today, many studies emphasize the role of transvaginal sonographic diagnosis of the female genital tract as a first line diagnostic test rather than the traditional laparohysteroscopy. A simple, ultrasound-based approach to investigate the infertile patient can be used effectively as an initial examination modality during the couple's workup [19]. A regular myometrial-endometrial interface and homogeneous endometrial structure on transvaginal sonography congruent with the phase of the menstrual cycle indicate a normal endometrium and preclude the need for diagnostic hysteroscopy. Transvaginal sonography may be used as the initial diagnostic procedure to select patients for hysteroscopy [20].

A review of 19 cases of documented retained fetal bones shows that diagnosis was made by uterine curettage in 12, hysteroscopy in 6 and hysterectomy in 1 patient. Six successful pregnancies were reported in this analyzed group of 19 patients following removal of retained fetal bones [7, 8, 10, 13, 14, 15, 16, 17, 21].

On reviewing the literature, the diagnosis of retained fetal bone was not discovered until hysterectomy in some patients. In others, it was discovered by a "gritty feeling" to the endometrium on D+C for abnormal bleeding and confirmed by pathology. In some cases, the fetal bone fragments were discovered on D+C after a filling defect was visualized on hysterosalpingogram. Bone fragments have been directly visualized protruding into the endometrial cavity on hysteroscopy in more recent cases (Figs. 1, 2, 3, 4). Occasionally, an echogenic area in the endometrium on transvaginal sonography may be the only clue to the presence of fetal bone fragments (Figs. 5, 6, 7).

A large secondary infertility case series by a Korean group [22] describes ten women with infertility of 1–5 years' duration after a therapeutic abortion. All of these women had a linear echogenic density discovered on vaginal ultrasound. After a D+C to remove the bone

Fig. 2 Hysteroscopic view of retained bone fragments, most probably the calvarium

Fig. 4 Hysteroscopic view of retained bone fragments, shaft of femur

Fig. 3 Hysteroscopic view of retained bone fragments, most probably cartilage

Fig. 5 Transvaginal sonogram (*sagittal view*) outlining a normal sized anteverted uterus with a hyperechogenic shadow in the region of the endometrial cavity suggestive of calcified intrauterine foreign body, which is a retained fetal bone in this case

fragments (three women required more then one curettage), all the women with open tubes conceived spontaneously and delivered healthy infants. Another recent case series with West African women [4] involved 11 women with secondary infertility of 2–15 years' duration. All these women had a previous pregnancy termination performed between 10 and 26 weeks gestation. Diagnosis of retained fetal bone was made with transvaginal ultrasound in all cases. Hysteroscopy was then performed to remove the bone fragments, and 8 out of the 11 women subsequently conceived spontaneously.

A blind D+C is sometimes diagnostically inaccurate and often ineffective as it may miss focal endometrial lesions. Chan [23] described how in his case the curettings from a blind D+C were normal before a hysteroscopy that visualized many retained fetal bone fragments. The best imaging technique that can readily visualize retained fetal bone is vaginal ultrasound. Any fetus that has attained at least 12 weeks gestation is capable of endochondral ossification. If that pregnancy is terminated or spontaneously aborted, any retained bone fragments might only be displayed on an ultrasound as bright echogenic areas with posterior shadowing (Fig. 6). It may not be adequate just to visualize the endometrium by hysteroscopy as some deeper endometrial pathology may be missed.

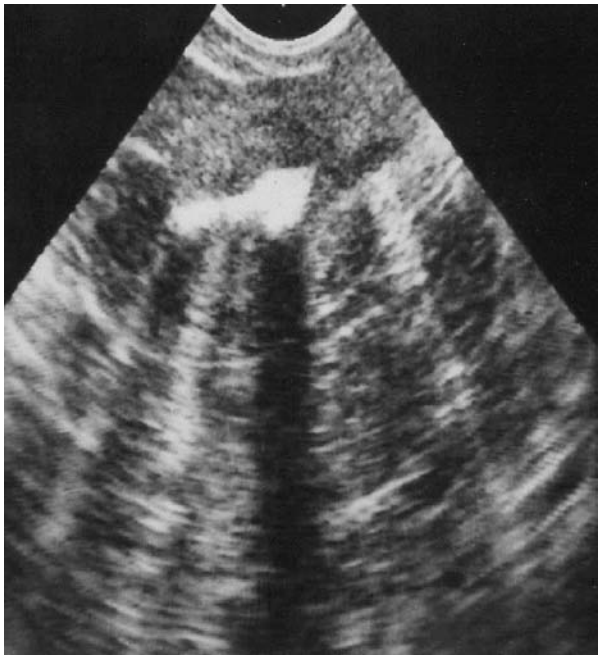

Fig. 6 Transvaginal sonogram (*coronal view*) showing the same retained bony fragments that cause an acoustic back scatter

Fig. 7 Naked eye view of two skull bones removed with a wire loop resectoscope

Discussion

Prolonged intrauterine retention of fetal bone parts is a now commonly diagnosed complication of induced abortion, spontaneous intrauterine fetal death and missed abortion. A case of long-term retention of fetal bone fragments in a 47-year-old Italian woman who underwent elective abortion 8 years earlier was reported by Panama et al. [24]. The patient was admitted for acute pelvic pain with purulent vaginal discharge. She reported recurrent episodes of abdominal and pelvic pain, meteorism, dysuria, nausea and vomiting, headaches and irregular cycles with dysmenorrhea and intermenstrual bleeding since the 1979 abortion. Initially, pelvic inflammatory disease was diagnosed and antibiotics were prescribed. When symptoms persisted after 10 days of drug treatment, the patient underwent a laparotomy that revealed pyosalpinx with extensive pelvic adhesions. Total hysterectomy with bilateral adnexectomy was performed. The uterine cavity was found to be closely packed with fragments that were determined at stereomicroscopic examination to be pieces of fetal bone. The retained fragments are an ideal substrate for bacterial colonization, which can spread to the tubes and destroy the functional integrity of the reproductive apparatus [24].

Similar to intrauterine contraceptive devices, fetal bones in the uterine cavity prevent conception. A 33-year-old woman was referred for infertility of 5 years' duration. Her history included pregnancy terminations at 6 weeks' gestation in 1992 and at 16 weeks' gestation in 1993. Both abortions were performed by curettage in West Africa. Her menses were every 28 days with normal flow. Due to a suggestion of a filling defect in the uterus on hysterosalpingography, hysteroscopic examination was performed. At hysteroscopy, several fragments of fetal bones were present in the uterus. These appeared as white structures, some with a trabeculated pattern. They were removed until the entire uterine cavity was devoid of bony fragments. Histopathology examination confirmed the diagnosis. It is very likely that the findings were related to the woman's abortion in 1993 [25].

Two cases of prolonged intrauterine retention of fetal bones were presented by Melius et al. and show that antecedent abortion may, though uncommonly, play a role in current gynecologic complaints [7]. In these two cases, symptoms dated to antecedent abortions treated with D&C 13 years and 14 months before diagnosis, respectively. Complaints included secondary infertility, dysmenorrhea and dysfunctional uterine bleeding. Hysteroscopy was necessary to make the correct diagnosis of retained fetal bones. In both cases, hysteroscopic surgery using graspers and forceps to pull out the fragments was unsuccessful in removing all the bony fragments or relieving symptoms [7]. Though retained fetal bones are an uncommon cause of gynecological problems, these cases show the necessity of hysteroscopy for diagnosis of persistent gynecologic problems when intrauterine pathology is suspect. These cases also demonstrate that although hysteroscopy is extremely useful diagnostically, it may not be successful therapeutically if the resectoscope is not used by a well-trained surgeon.

A 22-year-old black woman presented with secondary infertility. After almost 1 year of attempted diagnosis, a hysteroscopy was performed and a white-appearing tubular structure, which was similar to the noncopper-bearing portion of an IUD, appeared to be embedded in the fundus. Removal was difficult and the removed segments proved to be bone. Results indicated there were bits of fetal bone partially embedded at the fundal portion of the uterus for 6 years after an induced abortion [1]. Curettage yielded several more bone bits; repeat hysteroscopy confirmed that the uterus was empty. Probably the

bones acted as a uterine synechia or an IUD and thus prevented pregnancy [1].

Dajani et al. described a case of a 26-year old woman, gravida 2, para 1, who presented with failure to conceive 18 months after removal of a Lippes Loop IUD worn for 12 months [26]. The IUD had been inserted following a miscarriage at 18 weeks gestation, in which vaginal evacuation was bloody and difficult. The patient had a normal menstrual cycle and normal blood loss. The pelvic ultrasonogram showed a persistent focus of intrauterine illumination, and hysterosalpingography revealed an irregularity in the right cornu. Results of laparoscopy were negative. Four thin plates of bone, each 10 mm in the long axis, were removed at an exploratory dilatation and curettage. Microscopic examination showed necrotic bony trabeculae and bone marrow in the intervening spaces. A pregnancy test was positive 3 months later, and the patient gave birth to a healthy female weighing 3.3 kg [26]. In both this and the similar reported case in the previous paragraph, the bone fragments were located in the fundus. The contraceptive action was possibly related to an increase of prostaglandin concentration in the fundal endometrium, which is a common site for blastocyst implantation.

Chervenak et al reported four patients with symptomatic intrauterine retention of fetal bones [8]. Presenting complaints included infertility, irregular vaginal bleeding, vaginitis and spontaneous passage of fetal bones. According to the authors, hysteroscopy was invaluable both in confirming the diagnosis and in achieving successful removal of fetal bones. Zoricicic et al. reported on removing a diaphysis of a fetal bone 22 mm long from a 49-year-old patient with irregular and painful menstruation [27]. Two cases of endometrial ossification with secondary infertility were reported by Marcus et al.; pelvic ultrasonography suggested an intrauterine foreign body in one case [28]. Hysteroscopy was necessary to make the correct diagnosis and to remove the bony fragments from the two cases. One patient conceived naturally after the bony fragments were removed [28]. Hanssen and Rasmussen reported a case of menorrhagia caused by retained fetal bones after a missed abortion in the second trimester [29]. By hysteroscopy, several bone fragments were removed. The patient got pregnant 2 months later [29].

Bacceci et al. presented a case report with a very rare and long-standing cause of infertility, osseous metaplasia of the endometrium, and described its successful management by hysteroscopy [30]. A woman with a history of induced abortion 12 years ago failed to conceive from that time on. The infertility work-up was unrevealing except for the presence of intracavitary calcification on ultrasonography. After diagnostic hysteroscopy, solid bony spicules covering the posterior wall of the endometrial cavity were removed by wire loop resectoscope. Histology established the diagnosis of osseous metaplasia of the endometrium. The patient conceived in her second spontaneous cycle. Hysteroscopy was an effective means of extracting this heterotopic tissue, thereby re-establishing fertility even after a long period of infertility [30].

Recognition of foreign bodies in the uterine cavity is of great importance for successful treatment of secondary infertility: e.g., a piece of intrauterine contraceptive (after a removal failure), suture after cesarean section, or cylindrical and flat small bones after abortions. Kazakov et al. reported 69 foreign bodies that were removed with a wire loop resectoscope during 1992–1993 [31]. Sixty-one of the foreign bodies were intrauterine devices. The remaining eight women (11.9%) underwent fetal bone removals. Transcervical resection of foreign bodies (TCRF) is safe and efficient. Sufficient cervical canal distension, selection of equipment and methods to be used are important for successful TCRF [31]. As a non-invasive and effective monitoring method, B mode ultrasonography is the first choice to monitor for TCRF. In the initial few cases, till the hysteroscopic surgeon garners sufficient experience with the wire loop resectoscope, a simultaneous trans-abdominal B mode sonographic monitoring is advisable. This simultaneous sonographic visualization of the uterine cavity enhances the confidence of the operating surgeon. For patients with high risk factors for uterine perforation, laparoscopic monitoring can be done simultaneously. Laparoscopic ultrasonography monitoring has both the advantages of B ultrasonography and laparoscopy monitoring, but is invasive and expensive [32].

Our group reported a case of a patient with secondary infertility who had bits of fetal skull bone partially embedded all along the endometrial cavity for 7 years after an aspirotomy done for missed abortion [5]. Mrs. S. P., a 28-year-old patient, gravida 1, para 0 with one missed abortion (aspirotomy in January 1989), presented on 3 July 1995 with the complaint of secondary infertility for 7 years. She had previously been seen in two other clinics for irregular menstruation since her missed abortion 7 years ago. Menarche was at 13 years of age, and menstrual periods had been regular and normal until her aspirotomy done for missed abortion. In that confinement she was first examined at 7 months amenorrhea and was referred for an urgent USG because the uterus was much smaller than per period of gestation. USG revealed a missed abortion corresponding to 16 weeks gestational size. An elective aspirotomy was then performed upon her. After her aspirotomy she was suffered from menometrorraghia with spotting/bleeding free intervals of 10–12 days. She had been treated repeatedly over the last 7 years with hormonal therapy. She had undergone a D+C 4 years ago, and the histopathology revealed proliferative endometrium. She presented on 3 July 1995 with severe bleeding, and a diagnostic cum therapeutic curettage was performed upon her on 4 July 1995, which again revealed a histopathologic diagnosis of proliferative endometrium. A diagnosis of dysfunctional uterine bleeding secondary to anovulatory cycles was made, and it was planned to give her a single cycle of ovulation induction to check if she responded to clomiphene citrate. Semen analysis of the male partner was normal. She was referred for routine follicular studies on day 8 of her subsequent menstrual cycle following ovarian stimulation with 50 mg clomi-

phene citrate administered from 02–06 (Siphene, Serum Institute of India Limited, Bombay, India). Transvaginal sonography was performed, and the pelvic scan revealed a normal-sized anteverted uterus, which measured 6.3×5.4 x 4.6 cm. Both the ovaries were normal in size, position and echotexture and revealed multiple small follicles. On sagittal view (Fig. 5), the area of the endometrial stripe revealed a hyperechogenic shadow that extended all along the uterine cavity. The endometrial morphology could not be made out. On coronal view (Fig. 6), the scan again revealed a hyperechogenic shadow with acoustic backscatter in the region of the endometrial cavity. This was suggestive of calcified material in the uterine cavity. A diagnosis of prolonged intrauterine retention of fetal bones after abortion was made, and the patient was posted for hysteroscopy 2 days later. The day following the transvaginal sonographic diagnosis, a plain X-ray of the pelvis was taken. The X-ray revealed a small indistinct radioopaque area in the region of the uterus. A diagnostic hysteroscopy was performed, revealing small (1–6 mm) bone fragments (supposedly skull bone), which were lightly embedded all over the cavity, extending from the fundal region, both periosteal regions and the lower part of the uterine cavity. A resectoscope (Karl Storz GmBH, Tuttlingen, Germany and Chimco Biomedical, Bombay, India) was introduced, and the fragments were dislodged with gentle maneuvering and were removed under hysteroscopic video control. An endocamera (DX-CAM, Karl Storz GmBH, Tuttlingen, Germany and Chimco Biomedical, Bombay, India) was used to monitor the entire procedure, which was recorded on videocassette for documentation purposes. The hysteroscope was reintroduced to document that the cavity was clean after attempts at bone removal were completed. A total of 11 small (1–6 mm) bone fragments (supposedly skull bone) (Fig. 7) were removed and sent for histopathologic diagnosis, which confirmed our findings.

We have now removed retained fetal bones using the wire-loop resectoscope in 52 cases over 2 years at our tertiary care infertility management center (unpublished observations). Gonadotropin-releasing hormone analogues (GnRHa) have many new uses in gynecology [33, 34]. Several clinical studies have demonstrated a mean decrease in uterine volume after about 3–6 months of therapy [35]. These patients might be candidates for GnRHa therapy so that a smaller corpus with an atrophic endometrium might aid in hysteroscopic retrieval of the retained bony fragments. We have successfully tried this approach in 21 patients where we diagnosed retained intrauterine bone fragments on transvaginal sonography. On day 1 of their menses, each received 3.75 mg Goserelin Acetate Implant (Zoladex, Astra Zeneca, India); hysteroscopic removal of bone fragments was planned roughly 15–20 days following this injection. Our preliminary impression is that this preoperative drug therapy really helps us remove these retained bones far more easily than in the non-prepared cases. Figures 8, 9, 10, 11, 12 are some of the intraoperative pictures using a wire loop resectoscope.

Fig. 8 Hysteroscopic view of the uterine cavity showing the resectoscopic loop and a close-up view of one of the bony fragments

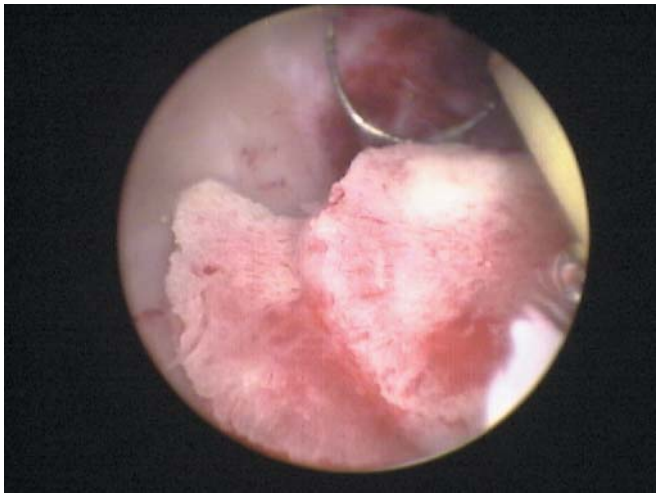

Fig. 9 Hysteroscopic removal of a retained bone piece using the wire loop resectoscope

Conclusions

Retained fetal bones should be considered in all patients with infertility, dysfunctional uterine bleeding, dysmenorrhea or other symptoms dating from a pregnancy or pregnancy termination [36, 37, 38, 39, 40]. The need to perform transvaginal sonography in all women who present with secondary infertility who had a previous pregnancy that progressed beyond 11 weeks gestation and then ended in spontaneous or therapeutic abortion is mandatory [36]. Prolonged retention of intrauterine bone is a recognized cause of secondary infertility. Transvaginal sonography is an excellent modality for evaluating these patients. Transvaginal sonography is also very useful for the follow-up of patients after surgical removal of the bony fragments. Some bony fragments may be embedded in the endometrium or myometrium and may not be identified at curettage. The simplicity of the treatment

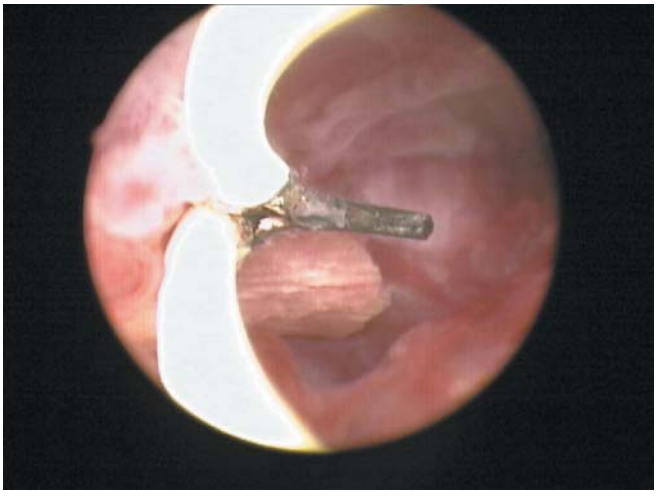

Fig. 10 Hysteroscopic view of the uterine cavity showing the resectoscopic loop and a close-up view of one of the bony fragments

Fig. 11 Hysteroscopic removal of a retained bone piece using the wire loop resectoscope

and the good postoperative prognosis justify an accurate and complete transvaginal sonographic examination of the endometrium in all women with secondary infertility who have a history of abortion. Not only will this examination identify unusual endometrial pathology, it may identify uterine anomalies that may have been missed previously. A diagnostic hysteroscopy followed by an operative procedure using a wire loop resectoscope is invaluable in both establishing the diagnosis and the removal of bony fragments. A crucial aspect of the procedure involves reintroduction of the hysteroscope to document that the cavity is clear after attempts at bone removal are complete. After removal of bony fragments, restoration of fertility and improvement of symptoms are expected.

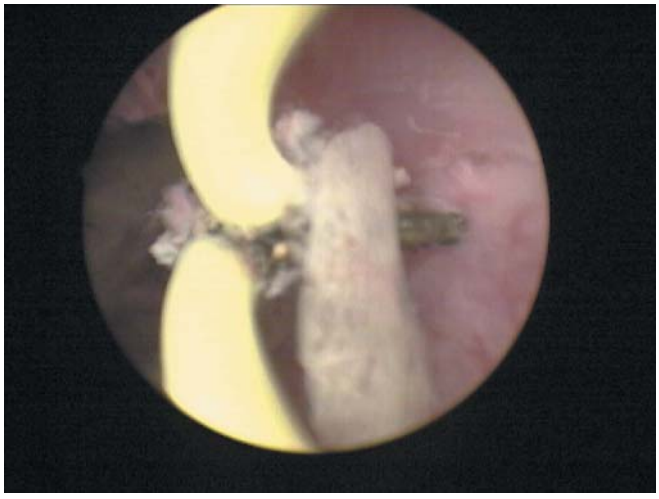

Fig. 12 Hysteroscopic view of the uterine cavity showing the resectoscopic loop and a close-up view of one of the bony fragments

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