International Surgery Journal
Anant M et al. Int Surg J. 2017 Oct;4(10):3573-3575
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: http://dx.doi.org/10.18203/2349-2902.isj20174544

Case Report

Osseous endometrium presenting as menorrhagia, successful outcome by hysteroscopy: a case report and review

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Received: 11 August 2017
Accepted: 07 September 2017

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ABSTRACT

Osseous endometrium is a rare clinical entity with the presence of bone in the endometrium. Almost all cases in literature have been reported as presenting with secondary infertility often having a prior history of a 1st or 2nd trimester abortion. This interesting case was 38 years age, presented with menorrhagia for few months only, had her last child birth 12 years ago by caeasarean delivery, her menstrual problem subsided with successful hysteroscopic removal of the bony chips. Although rare, the diagnosis is suspected on ultrasonography and can be managed easily with hysteroscopy which can visualize and simultaneously remove the bony spicules.

Keywords: Bone, Endometrium, Hysteroscopy, Infertility, Menorrhagia

INTRODUCTION

Osseous endometrium is a rare clinical entity with the presence of bone in the endometrium. It is rare and has mostly been misdiagnosed. It has been reported in literature mostly as cases of secondary infertility ranging from 1-14 years, often having a prior history of an abortion. The examination findings of these patients are usually unremarkable but sonography gives a clue by hyperechoic shadows in endometrium. The diagnosis clincher is direct visualization of the bony material by hysteroscopy and its removal almost magically results in fertility restoration. Menorrhagia (AUB-HMB) due to osseous changes in endometrium has not been reported in literature.

CASE REPORT

A 36-years old P1+ 1 L1 with history of cesarean section 12 years ago and a dilatation evacuation for an early 2nd trimester miscarriage 14 years ago, presented with heavy menstrual bleeding for 4 cycles. The cycles were regular but increased both in duration and flow. There was no dysmenorrhea, intermenstrual bleed or post coital bleeding. On examination mild pallor was present, abdominal and pelvic examinations were normal. Per speculum examination revealed normal looking cervix, Pap smear was reported negative for intraepithelial lesion or malignancy. Clinically uterus was normal sized with regular size, contour and non-tender, fornices were free. The patient had no signs or laboratory findings suggesting a calcium metabolism disorder. Ultrasonography was done and showed hyperechoic thickened endometrium (8.2mm) in the lower end of body of uterus, whereas the endometrium at the fundus. The dimensions of uterus were 10x3.7x3 cm and bilateral ovaries were normal. There was no free fluid in pouch of douglas.

A differential diagnosis of tuberculous endometrium, osseous metaplasia endometrium or least likely to be retained fetal bony parts (a cesarean full-term delivery had taken place after the second trimester evacuation), was made. Hysteroscopic evaluation of endometrial

cavity was undertaken which showed multiple white particles of fish lacy net appearance, some were floating in distension fluid and some embedded in posterior uterine wall at the level of internal os Removal of all these were done meticulously with the help of hysteroscopic forceps. The rest of the endometrial cavity had normal appearance of endometrial lining and ostia. Endometrial biopsy was also performed. Histopathology of the specimen reported presence of bone trabeculae in endometrium comprising normal stroma and glands. There was no surrounding tissue reaction.

DISCUSSION

Chronic inflammatory reaction in the endometrium may evoke metaplastic changes in the pluripotent endometrial stromal cells into osteoblastic cells that lay down bone.[1] Ossification of the endometrium is an uncommon clinical entity. It has been described by other names also such as endometrial ossification, ectopic intrauterine bone, and heterotopic intrauterine bone.[2,3] Ossification has also been reported in the cervix, the ovary and the vagina.[4-7]

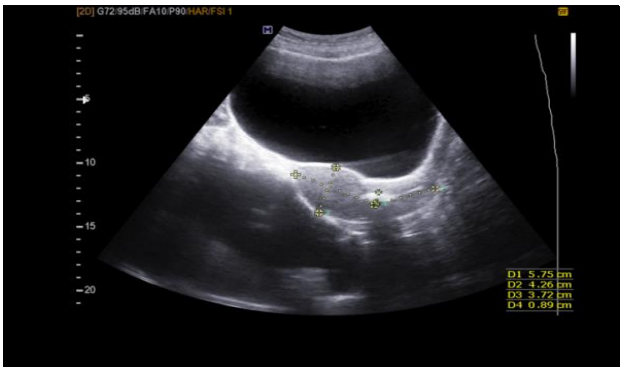

Figure 1: TAS image showing hyperechoic shadow in lower uterine segment.

Common clinical presentations of this entity are menstrual irregularities, pelvic pain, dyspareunia, vaginal discharge, and secondary infertility. Secondary infertility has been the most reported symptom of this disease, menstrual irregularities is least common. Majority of the patients are in the reproductive age group with history of first or second trimester abortion, either therapeutic or spontaneous and have normal menstrual cycle in the post-abortive period as noted in our case.[1-3,8,9] The time interval between the antecedent abortion and discovery of endometrial ossification varies from 8 weeks to 14 years. Osseous endometrium is an endogenous non-neoplastic pathological condition as no tissue reaction is found in the endometrial tissue studied and the endometrium showed normal regular cyclical changes in response to ovarian hormones.[1,3]

Various theories have been described in the published literature in the pathogenesis of endometrial osseous metaplasia. Ossification of post-abortive endometritis can be the first and most likely explanation for most cases detected after a preceding episode of miscarriage.[8]

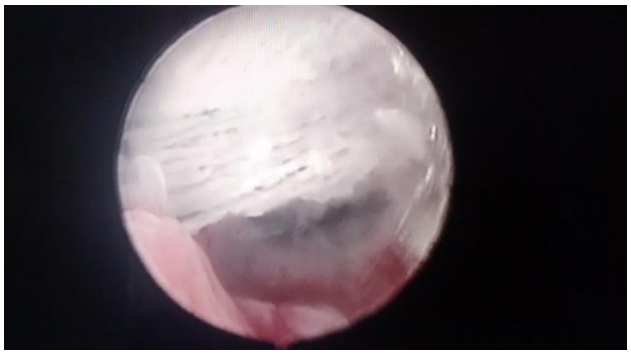

Figure 2: hysteroscopic view of uterine cavity showing embedded fish net bony spicule.

Figure 3: Bony chips removed by hysteroscopic grasper.

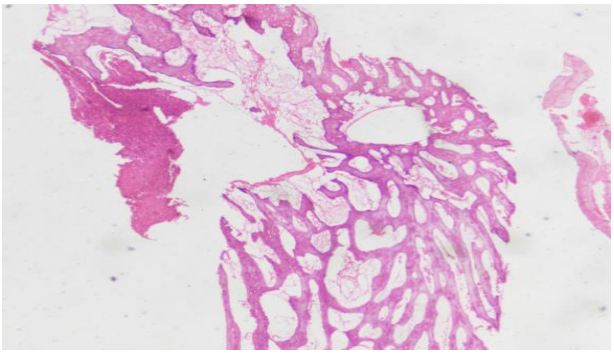

Figure 4: Histopathological image of spicules of bone embedded in endometrial glands and stroma.

The other hypothesis of origin of osseous material in endometrium can be due to presence of inflammation (acute/chronic endometritis) which can induce metaplastic changes in endometrium. This is more likely in nulliparous patients. In India awareness of tuberculous pathology in genital tract and chronic inflammation due to this may cause calcifications (dystrophic calcification/ metastatic calcification) which can give appearance as hypoechoic in ultrasonography. Retained fetal bone has also been proposed as one of the theories. There can also

be metabolic disorders of calcium and vitamin D as reported by some.[1-3,9,10] The diagnosis can be suspected by ultrasonography but there can be confusion with intrauterine contraceptive device as both present the same image characteristics. The treatment before advent of hysteroscopy was blind dilatation and curettage which used to be associated with either incomplete removal or excessive/vigorous curettage. In either case the end result was persistence of infertility in most women.

Ultrasound guided dilatation curettage was the next step forward, which could give some better evacuation of the uterine cavity and successful removal of most osseous material. The direct visualization and removal of osseous material by hysteroscopy is the current modality of diagnosis and treatment as well. Various reports of successful management of osseous endometrium by hysteroscopy are there in literature.[9,11] Removal has led to successful pregnancy outcomes in most women presenting with secondary infertility. Bozdag et al have reported that restoration of endometrial cavity with hysteroscopy or curettage provides a spontaneous pregnancy rate of 54.2% within 12 months.[12] They also recommended that irrespective from the duration of subfertility, assisted reproduction cycles should be postponed, unless there is another reason for infertility. That spontaneous pregnancy can be expected within a year of normal restoration of endometrial cavity. For those who do not conceive, assisted reproductive techniques may help.[13]

In a large systematic review of 216 patients it has been reported that bone fragments in the endometrium are most commonly found after pregnancy termination, present with infertility and/or irregular menses, and upon removal, patients rapidly conceive spontaneously.[14] Menorrhagia due to osseous endometrium can be due chronic inflammation leading to endometritis or release of prostaglandins like PGE2 vasodilates the vessels of the endometrium, and PGI2 relaxes smooth muscle, vasodilates the vessels of the myometrium and inhibits thrombocyte aggregation.[15] PGF2 alpha vasoconstricts the endometrial vessels during menstruation and contracts the smooth muscle of the myometrium. The pathological conditions of dysmenorrhea and menorrhagia are probably due to increased PG levels.

CONCLUSION

Menorrhagia is caused due to a number of causes but finding of osseous material is a rare cause. Imaging can help but the method of choice for diagnosis and treatment remains hysteroscopy which can visualize directly and remove at one sitting, it is safe and simple method and also confirms complete removal and restoration of normal endometrial cavity.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Anant M, Singh A. Osseous endometrium presenting as menorrhagia, successful outcome by hysteroscopy: a case report and review. Int Surg J 2017;4:3573-5.