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An unusual case of adenomyosis reported in Central India

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Abstract

Adenomyosis is the presence of endometrial glands and stroma within the myometrium. A 40 years old nullipara presented with complaints of heavy bleeding during menses with passage of clots and white discharge with pain in abdomen more during menses for 3 years. On per abdomen examination a globular mass seen in lower abdomen below umbilical. It was about 20 weeks uterus size, firm, smooth with regular margins. Ultrasound was done which suggested bulky uterus of 9.6x11.9x8.5 cm, with heterogeneous myometrium globular fundus with increased vascularity and indistinct endomyometrial junction. Endometrial thickness of 4mm and normal ovaries. Mild hydronephrosis on both sides. MRI done suggested solid vascular mass lesion of 14.1x12x5.2cm with smooth margin in posterior wall of uterus extending to anterior wall of fundus with multiple cystic degeneration. Patient was not desirous of child in future and wanted permanent cure. So total abdominal hysterectomy with bilateral DJ stenting was done. Diffuse Adenomyosis was eventually confirmed in histopathology report.

Keywords: adenomyosis, myometrium, hysterectomy

Introduction

Adenomyosis is a condition that affects the uterus. In women with adenomyosis, the endometrial tissue (which typically lines the uterus) moves into the outer, muscular walls of the uterus. Some women may have no signs or symptoms of the condition. When present, features of the condition include heavy menstrual bleeding, painful menstrual periods, and pelvic pain during intercourse. Some women may also develop an adenomyoma, which is a mass or growth within the uterus. The underlying cause of the condition is currently unknown ^[1].

The modern definition of adenomyosis was provided in 1972 by Bird who stated: "Adenomyosis may be defined as the benign invasion of endometrium into the myometrium, producing a diffusely enlarged uterus which microscopically exhibits ectopic non-neoplastic, endometrial glands and stroma surrounded by the hypertrophic and hyperplastic myometrium" ^[2]

In simple words, Adenomyosis is the presence of endometrial glands and stroma within the myometrium.³

70 to 80% of women undergoing hysterectomy for adenomyosis are in their fourth and fifth decade of life and are multiparous; several studies have reported a mean age over 50 years for women undergoing hysterectomy for adenomyosis ^[4-13]

A high percentage of women with adenomyosis are multiparous ^[8-10, 14, 15].

Case Report

Here we report a case of unusual presentation of adenomyosis in a 40-year nullipara with 20-week size uterus not associated with fibroid.

A 40 years old nullipara presented with complaints of heavy bleeding during menses with passage of clots and white discharge with pain in abdomen more during menses for 3 years. Patient also had lump in lower abdomen noticed 1 year back. Menstrual cycles were regular, bleeding continued for 8- 10 days. There were no urinary complaints. Patient was previously given Gonadotropin releasing hormone (GnRH) agonist but was not relieved. She never conceived and did not take any treatment for the same. There was no past surgical history. Family history was insignificant.

On examination patient was vitally stable and clinically pale. On per abdomen examination a globular mass seen in lower abdomen below umbilical. It was about 20 weeks uterus size, firm, smooth with regular margins, lower margin could not be reached, freely mobile transversely and

limited mobility upside down. Per speculum examination suggestive of grossly healthy cervix, pulled up with minimal white discharge. On Per-vaginal examination same mass could be palpated, mobile with movement of cervix.

Ultrasound was done which suggested bulky uterus of 9.6x11.9x8.5 cm, with heterogeneous myometrium globular fundus with increased vascularity and indistinct endomyometrial junction. Endometrial thickness of 4mm and normal ovaries. Mild hydronephrosis on both sides.

MRI done suggested solid vascular mass lesion of 14.1x12x5.2cm with smooth margin in posterior wall of uterus extending to anterior wall of fundus with multiple cystic degeneration suspecting Adenomyoma.

Patient was not desirous of child in future and wanted permanent cure. So total abdominal hysterectomy with bilateral DJ stenting was done. Ovaries were healthy and preserved. Uterus was 22x16x16cm, symmetrically enlarged with 1.5kg weight.

On cut section no localized mass could be seen, myohyperplasia observed in walls with cystic degeneration and indistinct endomyometrial junction.

Diffuse Adenomyosis was eventually confirmed in histopathology report.

Discussion

This case report brings light to the fact that a lot more needs to be discovered and studied about adenomyosis. Adenomyosis can present in unique ways. As the causation of adenomyosis is linked with breach in endomyometrial junction due to trauma during delivery or any intervention, which results in internalization of endometrial glands. In this case no cause for breach in endomyometrial junction was observed. It favours theory of genetic predisposition. According to Parrot *et al* nerve growth factor-alpha, preadipocyte factor-1, and insulin-like growth factor-2 genes may play an important role in regulating differentiation and development of the myometrium, these data suggest that adenomyosis may be caused primarily by defects in the formation of the myometrium [16].

This case is an exception to the fact that adenomyosis uterus does not grow beyond 14weeks size. Over 14 weeks growth is associated with fibroid in about 60% cases. In this case uterus symmetrically enlarged to 20 weeks size with 1.5kgs weight. Cut section shows granular pattern with no circumscribed area with capsule, denying coexistence of fibroid. This case is of severe adenomyosis according to Bird *et al* grading and grade III by Molitor's criteria [17, 18] Based on depth it is Deep Adenomyosis.

The accuracy of investigation is decreased in women with coexisting fibroid or focal adenomyosis. It may be misdiagnosed in ultrasound as it may be taken as multiple leiomyoma or endometrial thickening. Pathological confirmation can be made at the time of hysterectomy. Histopathology remains confirmatory for diagnosis.

Adenomyosis is less commonly associated with infertility. Its role in infertility is still debatable but frequency of infertility has not been assessed. It has been seen to be associated with pregnancy which favors invagination of nasal endometrium into myometrium.

Adenomyosis unlike endometriosis, is refractory to hormone treatment. GnRH agonists are efficient in reducing the adenomyosis uterus size and may facilitate fertility. Recurrence of adenomyosis is common after discontinuation of GnRH agonists. Conservative surgical treatment is available but hysterectomy holds the principal diagnostic, therapeutic and definitive treatment for deep adenomyosis.

Conclusion

This case draws attention to rare presentation of adenomyosis. Association with infertility, growth beyond 14weeks uterus size without association with fibroid and newer theory of causation. MRI is superior to ultrasound in diagnosis of adenomyosis. GnRH agonists have limited role in providing relief in deep adenomyosis. Hysterectomy is still the definitive treatment of choice in such a case.

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