



RESEARCH ARTICLE

AN UNUSUAL CASE OF FOCAL ADENOMYOTIC CYST DIAGNOSED AND MANAGED HYSTEROSCOPICALLY

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ABSTRACT

Focal adenomyotic cyst is defined as a cystic structure lined by endometrial tissue and surrounded by the myometrium. It is a rare and benign pathology. It is seen in adolescents and young women of reproductive age group. The presenting complaints are non specific. Most commonly, women present with dysmenorrhea, chronic pelvic pain or abnormal uterine bleeding. Now a day's its incidence is rising, one because of increasing age of conception and second because of availability of high quality imaging modalities and minimally invasive techniques such as hysteroscopy. With this background, author here report a case of 29 years old woman who presented with primary infertility since 7 years. Her laboratory investigations were normal. Ultrasound pelvis was normal. Diagnostic hysteroscopy was done, which showed mild adhesions in the uterine cavity, for which fundal and lateral wall metroplasty was done. After adhesiolysis, a brown coloured cystic lesion was visualised on posterior wall of uterus, of size 2 × 2 cm, which was excised with 5 Fr scissors. On excision, haemorrhagic fluid was drained, thus confirming the diagnosis. On conclusion, although adenomyotic cyst is a rare form of adenomyosis but now since the incidence is on rising trend, thus gynaecologists need to be more vigilant for its diagnosis and management. Through this case, authors would like to highlight the diagnostic and therapeutic role of hysteroscopy for focal adenomyotic cyst.

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INTRODUCTION

Adenomyosis is defined as presence of endometrial glands and stroma in the uterine myometrium. It can be diffuse or focal. Diffuse adenomyosis is the most common form of adenomyosis while focal adenomyosis in the form of adenomyotic cyst is rarely seen. The focal adenomyotic cyst is defined as a cystic structure lined with endometrial tissue and surrounded by myometrial tissue. It is a rare and benign pathology. Cucinella *et al* in 2013 (Cucinella *et al.*, 2013) has reported in their article that till date only 30 reports of such adenomyotic cysts with similar characteristics has been found in the literature. Diffuse adenomyosis is primarily a disease of elder age group while focal adenomyotic cyst is seen commonly in adolescents and women younger than 30 years (Brosens, 2015). The presenting complaints are non specific. Most commonly, women present with dysmenorrhea, chronic pelvic pain or abnormal uterine bleeding. Overall, it is a rare cause of abdominopelvic pain and dysmenorrhea in adolescents and young women (Ho *et al.*, 2009).

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Traditionally adenomyosis has been diagnosed by pathologist in hysterectomy specimens; however recent development of high quality imaging modalities and minimally invasive techniques such as hysteroscopy has enabled the clinicians to diagnose it in the office setting. Diagnosis is primarily based on 3-D ultrasound and magnetic resonance imaging on which it appears as a cystic structure with hemorrhagic content surrounded by myometrial tissue. In recent years, Hysteroscopy is being used as a diagnostic tool for evaluation of uterine cavity (Dobashi, 1992 and Steinkampf *et al.*, 2004). On hysteroscopy, adenomyotic cyst is diagnosed as a submucous cyst bulging into the uterine cavity, and visible clues such as abnormal vascularisation or fibrosis, may be observed in the endometrium at the site of the cyst. Hysteroscopic excision can also be done at same sitting either by scissors or by ablation. With this background, the authors would like to discuss case of focal adenomyotic cyst.

Case Report

A 29 years old nulliparous woman presented to our outpatient department with complaint of inability to conceive since 7 years. She was having regular menstrual cycle at interval of 30

days, lasting for 5 days, with average blood flow. There was no complaint of dysmenorrhea or chronic pelvic pain. She underwent laparoscopic surgery 2 years back, in which bilateral ovarian cystectomy was done. After the surgery, IUI was done twice but unfortunately, she did not conceive. Work up for infertility was done. Her blood investigations were normal. Hormonal investigations were normal. Husband semen analysis was normal. Ultrasound pelvis was done, which showed uterus of normal size and shape without any space occupying lesion. Bilateral adnexa were normal. She was being planned for IVF and before that diagnostic hysteroscopy was done. On hysteroscopy, she was found to have mild adhesions (Figure 1 A), for which adhesiolysis was done with 5 Fr scissors. After the adhesiolysis, a blue coloured cystic lesion of size 2 × 2 cm was visualised on the posterior wall of uterus, near the right lateral wall and right ostia (paramedian in location) (Figure 1 B). Further adhesiolysis was done to completely expose it. Excision was then carried out with 5 Fr scissors (Figure 1 C), following which brown coloured haemorrhagic fluid was drained (Figure 1 D), thus confirming the diagnosis of focal adenomyotic cyst.



Figure 1 A. A wide panoramic view of uterine cavity showing mild adhesions at fundus and lateral wall



Figure 1 B. bluish coloured cystic lesion visualised at posterior wall of uterus after metroplasty



Figure 1 C. Excision of adenomyotic cyst with 5 Fr scissors



Figure 1 D: Drainage of haemorrhagic fluid after excision of adenomyotic cyst

DISCUSSION

Focal adenomyotic cyst is a rare and benign pathology (English, 2012). The first case has been described by Cullen *et al.* in 1908 (Cullen, 1908).

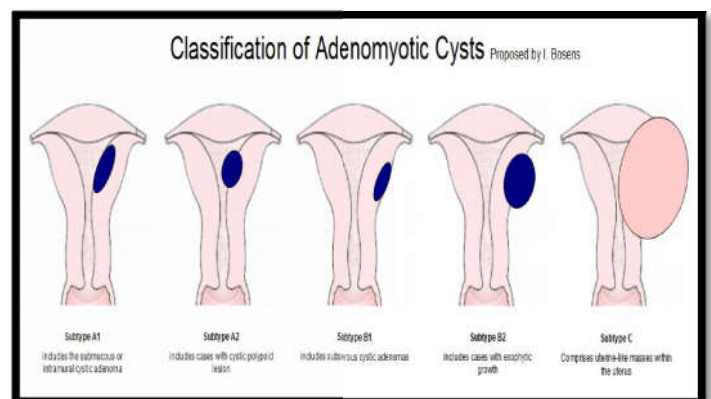


Figure 2. Figure showing classification of focal adenomyotic cyst as given by Brosens et al

However, now its incidence is rising probably because of the increasing age of conception and also availability of better imaging modalities to diagnose the pathology (Gordts, 2014). Various theories of pathogenesis of adenomyotic cyst have been given as invagination of endometrial tissue into the myometrial tissue or estrogenic stimulation of Müllerian remnants in the myometrium or iatrogenic implantation of endometrial tissue into the myometrium during uterine surgery (Ferenczy, 1998).

The clinical features of presentation are non specific. The most common symptom is dysmenorrhea, which starts at an early age, around the time of menarche, tends to increase progressively with age and is resistant to medical therapy including analgesics or cyclic oral contraceptives. Other symptoms include abnormal uterine bleeding, chronic pelvic pain and infertility. Brosens *et al.* (1993) reported an incidence of adenomyotic cyst as 50% in patients with sub fertility, dysmenorrhea, and dysfunctional uterine bleeding. An estimated prevalence of adenomyotic cyst in patients with endometriosis has been reported to be approximately 70% by Kunz *et al.* (2005). Since the introduction of imaging techniques, an increasing number of cases have been described in adolescents and young adult women. On 3 D ultrasound or MRI, it appears as cystic structure with an internal diameter of ≥ 10 mm, surrounded by myometrial tissue. However, adenomyotic cyst might not be visualized on ultrasound and may appear as sub endometrial haemorrhagic area, thus get missed on these techniques.

Table 1. Table showing signs of adenomyotic cyst on hysteroscopy

Pathognomic signs of adenomyotic cyst on hysteroscopy
1. Submucous adenomyotic cyst
2. Irregular endometrium with endometrial defects
3. Altered vascularisation
4. Cystic hemorrhagic lesion

The differential diagnosis of uterine intramural cystic lesions includes non-communicating rudimentary horn, cystic degeneration in a leiomyoma and adenomyosis. Hysteroscopy is an emerging diagnostic as well as therapeutic tool for adenomyotic cyst. It is visualised as a cystic structure bulging into the uterine the cavity. Lowering the intrauterine pressure is helpful for a better identification of the sub mucosal cystic structures. However, when sometimes diagnostic hysteroscopy is not able to reveal the pathognomonic signs of adenomyosis, then some studies suggests that visible clues (Molinas, 2006) should be considered for its diagnosis including an irregular endometrium with endometrial defects, altered vascularisation, and cystic hemorrhagic lesion, as shown in Table 1. The adenomyotic cyst is classified as different subtype A1, A2, B1, B2, C as given by Brosens *et al.* (1993), (Figure 2). Surgical resection of cyst can be done by various methods. In cases of submucous cystic adenomyotic lesions, direct hysteroscopic access is possible. Mechanical excision can be done by using 5 Fr scissors. Using 5-Fr scissors during hysteroscopy allows a clear dissection of the wall of the cyst from the surrounding myometrium. Another way of doing excision is by ablation via energy source. An ablative technique actually destroys the whole inner cystic wall of the adenomyotic cyst. Ablative approach is preferable for those cysts which are localized

deeper in the intramural portion. In cases in which intramural cystic structures are present, ultrasound guidance is preferable for localization of the cystic structure.

A case report of adenomyotic cyst was done in 2005, in which a 46 year old woman was found to have an anechoic area on transvaginal ultrasound, hysteroscopy was done, which revealed a cystic mass on the posterior wall of uterus, which was excised by means of a bipolar loop resectoscope (Giana, 2005). Another case was published in 2006, of a 21 year old woman who had severe dysmenorrhea, supposedly caused by cystic adenomyosis. Excision of cyst was done with radio-frequency needle inserted via the cervix under ultrasound guidance under general anesthesia. This was the first case reported as adenomyotic cyst excised by radiofrequency ablation (Ryo *et al.*, 2006). In 2014 a study was published describing 2 cases of adenomyotic cyst, out of which one case presented with menorrhagia and she was found to have adenomyotic cyst on hysteroscopy. Excision of cyst was done with 5 Fr scissors. Another case was of 38 years old woman who presented with primary infertility, who was a diagnosed case of adenomyotic cyst. Hysteroscopic resection was done by using bipolar resectoscope (Gordts *et al.*).

Conclusion

Although adenomyotic cyst is a rare form of adenomyosis but now a day its incidence is increasing due to delayed age of conception and availability of accurate imaging techniques. Hysteroscopy is a diagnostic tool for visualisation of uterine cavity with a direct access to Adenomyotic cyst. Hysteroscopy also allows the simultaneous treatment of adenomyotic cyst via excision or ablation.

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