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RESEARCH ARTICLE

HYSTEROSCOPY: A DIAGNOSTIC & THERAPEUTIC TOOL FOR FOCAL ADENOMYOTIC CYST

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ABSTRACT

Introduction: Adenomyosis is defined as presence of endometrial glands and stroma in the uterine myometrium. It can be diffuse or focal. Diffuse form is commonly seen while focal adenomyotic cyst is a rare entity. Recent development of high quality imaging modalities and minimally invasive techniques such as hysteroscopy has enabled the clinicians to diagnose the disease in the office setting. Hysteroscopy not only help in its diagnosis but also its simultaneous management.

Case series: Authors here are presenting a case series of focal adenomyotic cyst diagnosed hysteroscopically over a period of eight years. Total 1173 women underwent hysteroscopy over this time period, of which nine cases of adenomyotic cyst were found. Of these nine women, six presented with primary infertility, one presented with secondary infertility and two of them came with complaint of abnormal uterine bleeding. Range of age presentation was 23-35 years. Among these nine cases, hysteroscopy revealed adenomyotic cyst which was excised at the same sitting with either 5 Fr scissors or resectoscope.

Conclusion: On conclusion, adenomyotic cyst is a rare form of adenomyosis with incidence found here is 0.76%. However now-a-days, more women are being diagnosed with adenomyotic cyst due to advance age at first pregnancy and availability of better imaging modalities and minimally invasive techniques. Thus via these cases, authors would like to highlight the role of hysteroscopy in diagnosis and management of adenomyotic cyst. The authors hereby are reporting the first ever case series with nine such cases in this age group, all managed hysteroscopically.

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INTRODUCTION

Adenomyosis is defined as hetrotopic presence of endometrial glands and stroma in the uterine myometrium. It can be diffuse or focal. The focal adenomyotic cyst is defined as a cystic structure lined with endometrial tissue and surrounded by myometrial tissue. It is a benign and rare pathology. It is seen commonly in adolescents and young women of age less than 30 years [1].

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. In recent years, Hysteroscopy is being used as a diagnostic tool for evaluation of uterine cavity [2-4]. On hysteroscopy, adenomyotic cyst is diagnosed as a submucous cyst bulging into the uterine cavity, and visible clues such as abnormal vascularisation or bluish discolouration or fibrosis, may be observed in the endometrium at the site of the cyst which should raise the suspicion of diagnosis. With this background, the authors would like to discuss focal adenomyotic cyst via a case series of nine cases.

Cases

Authors here describe a case series of nine cases of focal adenomyotic cyst diagnosed over a period of eight years from January 2009 to May 2016. Total 1173 women underwent hysteroscopy during this time period, out of which 9 women were found to have adenomyotic cyst. Thus incidence of focal adenomyotic cyst is found to be 0.76%, showing the rarity of disease. The most common indication of doing hysteroscopy was infertility (54%) followed by abnormal uterine bleeding (28.1%) as shown in table I. Following are the case summaries of these nine cases of focal adenomyotic cyst.

Table I Table showing total number and percentage of women undergoing hysteroscopy with its indication.

Indication of hysteroscopy	Number of women	Percentage of women
Abnormal uterine bleeding	**331**	**28.2%**
Infertility	**727**	**62%**
Missed IUCD	**8**	**0.7%**
Amenorrhea	**10**	**0.8%**
Endometriosis	**41**	**3.4%**
Relook hysteroscopy	**41**	**3.4%**
Endometrial hyperplasia	**15**	**1.3%**
Total	**1173**	**100%**

- A 29 years old woman presented with primary infertility since 7 years. She was having regular menstrual cycles. There was no complaint of pelvic pain, dysmenorrhea, and discharge per Vaginum. She had history of laparoscopic bilateral ovarian cystectomy done 2 years back, endometriotic in origin. After the surgery, 2 cycles of IUI was done but unfortunately she did not conceive. Her blood and hormonal investigations were normal. Husband semen analysis was normal. Ultrasound pelvis was done, which was reported as normal. She was planned for IVF, before which diagnostic hysteroscopy was done. On hysteroscopy, uterine cavity was narrow in calibre, for which fundus and lateral wall metroplasty was done. Following adhesiolysis, a bluish coloured cystic lesion of size 2 × 2 cm was visualised on posterior wall of uterus (figure 1A). After doing adhesiolysis over the surface of lesion, an adenomyotic cyst was revealed (figure 1B), which was excised with 5 French scissors followed by which haemorrhagic fluid was drained. After the surgery, patient has achieved an IVF pregnancy.

Figure 1A Hysteroscopic image with bluish coloured lesion visualised on the posterior wall of uterus.

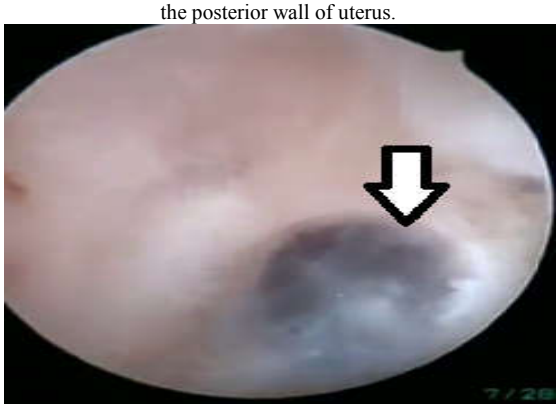

Figure 1B Hysteroscopic image with arrow pointing towards adenomyotic cyst on the posterior wall of uterus.

- A 26 years old woman, $P_1L_1A_1$ presented with secondary infertility. She had history of oligomennorhea since one year for which she was evaluated. Her blood and radiological investigations were normal. Diagnostic hysteroscopy was done, on which Asherman's syndrome was diagnosed and metroplasty was done. After 6 weeks, second look hysteroscopy was done, which revealed mild adhesions for which fundal and lateral wall metroplasty was done (figure 2A). A small elevated lesion was visualized on the posterior wall of uterus (figure 2B). Using mechanical 5 French scissors, adhesiolysis over that lesion was done, following which a bluish coloured small lesion of size 0.5 × 0.5 cm was seen (figure 2C). On excision, haemorrhagic fluid was drained (figure 2 D) thus confirming the diagnosis of adenomyotic cyst.

Figure 2 A Hysteroscopic image showing panoramic view of uterine cavity

Figure 2 B Hysteroscopic image after right lateral wall metroplasty, showing elevated area over posterior wall of uterus.

Figure 2 C Hysteroscopic image showing bluish coloured adenomyotic lesion after adhesiolysis over its surface.

Figure 2 D Hysteroscopic image with drainage of brown coloured haemorrhagic fluid after excision of adenomyotic cyst.

- A 33 years old woman presented with primary infertility. She had history of 3 failed intrauterine insemination cycles. She was having regular menstrual cycles. Her blood investigations and hormonal profile was normal. Male factor of infertility was also ruled out. Ultrasound pelvis was done which showed an anechoic cystic lesion of size 2 × 2 cm in uterus with normal ovaries. She was planned for IVF and pre IVF diagnostic hysteroscopy was done. On hysteroscopy, cervical canal was normal. Uterine cavity was found to have elevated lesion on the posterior wall of uterus (figure 3A). Adhesiolysis was done with resectoscope on posterior wall, following which haemorrhagic fluid was drained out from the lesion (figure 3B, C, D). Complete excision of adenomyotic cyst was done using resectoscope (figure 3C, D). Patient has conceived after surgery.

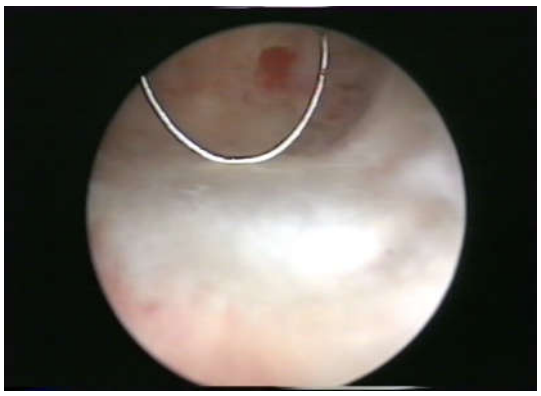

Figure 3A Hysteroscopic image showing elevated area on the posterior wall of uterus.

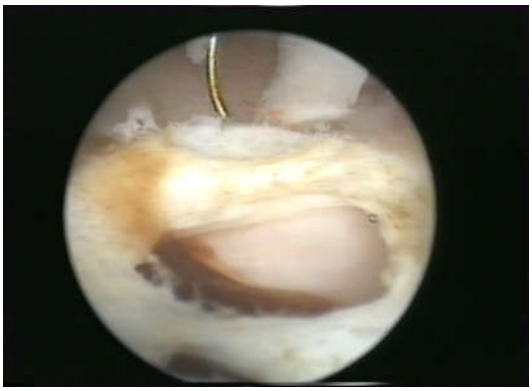

Figure 3B Hysteroscopic image showing drainage of haemorrhagic fluid after resection with resectoscope.

Figure 3C Hysteroscopic image showing further drainage of haemorrhagic fluid after deep resection of cyst.

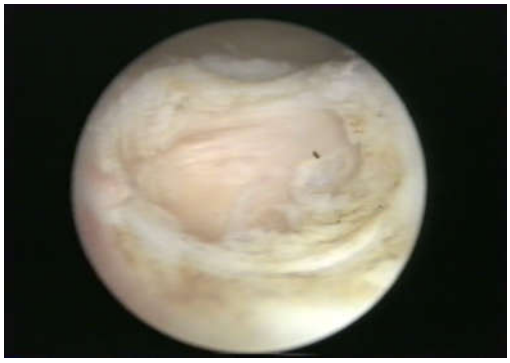

Figure 3D Hysteroscopic image showing complete excision of adenomyotic cyst on posterior wall of uterus.

- A 28 years old woman presented to our centre with primary infertility. There was no complaint of abnormal uterine bleeding, dysmenorrhea or discharge per Vaginum. Her blood investigation, hormonal profile, husband semen analysis, ultrasound pelvis was normal. Diagnostic hysteroscopy was done which revealed normal cervical canal with uterine cavity showing small 1 × 0.5 cm adenomyotic lesion on right lateral wall, which was excised with 5 Fr scissors.
- A 23 years old woman was referred to us with primary infertility for pre IVF hysteroscopy. She was married for 3 years, but unable to conceive. Her investigations for infertility were normal. Ultrasound pelvis was normal. She had received ovulation induction. 3 IUI cycles were done but she did not conceive. On hysteroscopy, uterine cavity was found to be subseptate, fundal metroplasty was done. Flimsy adhesions were seen on left lateral wall, adhesiolysis was done, and following which small brown coloured adenomyotic cyst was seen near left ostia of size 0.5 × 0.5 cm, which was resected out.
- 35 years old woman presented with severe dysmenorrhea and primary infertility. On ultrasound she was found to have bilateral ovarian cyst. With provisional diagnosis of endometriosis, she was planned for hysteroscopy with laparoscopy. On hysteroscopy, adenomyotic cyst of size 1 × 2 cm was visualised on posterior wall of uterus, which was excised with 5 Fr scissors. On laparoscopy, uterus was normal, bilateral endometriotic ovarian cyst was seen and bilateral cystectomy was done.
- Another 30 years old woman presented with primary infertility. She was married for 7 years. She was having normal menstrual cycles, without any complaint of dysmenorrhea. Her laboratory and imaging investigations were normal. On hysteroscopy, she was found to have small cavity. Adhesiolysis was done with Monopolar resectoscope. After adhesiolysis on posterior lateral wall, focal adenomyotic cyst of size 2 × 1 cm was visualised. Cyst was ablated, following which haemorrhagic fluid was drained.
- 25 years old woman presented with heavy menstrual bleeding since four months. She was having regular menstrual cycles of 28 days, lasting for 10-12 days, soaking 4-5 pads in a day, with passage of clots. On pelvic examination, cervix and vagina was healthy,

uterus was anteverted, mobile, normal size and bilateral fornices were free. Ultrasound examination was done, which was reported as normal. Diagnostic hysteroscopy was done, which revealed a focal adenomyotic cyst of size 2 × 2 cm on posterior wall. It was excised by using Monopolar resectoscope and haemorrhagic fluid was drained.

- A 32 years old woman presented with heavy menstrual bleeding. She had history of medical abortion 2 months back. Her blood investigations were normal. On ultrasound, endometrial polyp of size 2 × 3 cm with vascular pedicle was visualised. Provisional diagnosis of placental polyp was made and patient was taken for hysteroscopic polypectomy. On hysteroscopy, polyp was seen on right cornual end which was excised. After the excision, adenomyotic cyst was visualised of size 0.5 × 0.5 cm, (figure 4 A, B) which was excised with resectoscope, following which brown coloured haemorrhagic fluid drained.

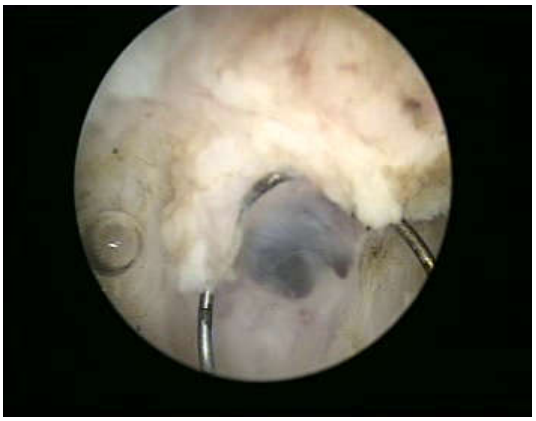

Figure 4A Hysteroscopic image with resection being done with resectoscope.

Figure 4B Hysteroscopic image showing bluish coloured adenomyotic lesion after resection.

DISCUSSION

Focal adenomyotic cyst is a rare and benign pathology [5]. Various names given to this pathology are adenomyotic cyst, cystic adenomyosis and cystic Adenomyoma.

The first case of focal adenomyotic cyst was described by Cullen TS in 1908 [6]. In 2013, in an article by Cucinella G *et al* [7], it has been reported that only 30 case reports of these cysts with similar characteristics has been reported till date. In 2010, Takeuchi H *et al* [8] had described a study of nine cases of juvenile cystic adenoma, where all cases were managed laparoscopically. The authors hereby are reporting the first ever case series of nine cases of focal adenomyotic cyst in women of reproductive age group, managed hysteroscopically. It is primarily seen in adolescents and women of reproductive age group. Although, earlier this entity was rarely seen however, now its incidence is on rising trend probably because of the increasing age of conception and also due to the availability of better imaging modalities to diagnose the pathology [9].

Various theories for pathogenesis of adenomyotic cyst have been given by Ferenczy A in 1998 [10]. One of the theories is invagination of endometrial tissue into the myometrial tissue leading to formation of cystic adenomyosis. Secondly, stimulation of Müllerian remnants in the myometrium by estrogens leads to development of adenomyotic cyst. Lastly, iatrogenic implantation of endometrial tissue into the myometrium during uterine surgery can also cause formation of focal adenomyotic cyst.

The clinical features of presentation are non specific. The various symptoms are dysmenorrhea, abnormal uterine bleeding, chronic pelvic pain and infertility. Of these, one of the most common symptoms 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. Brosens *et al*. in 1993 [11] in a study reported an incidence of adenomyotic cyst as 50% in patients with sub fertility, dysmenorrhea, and dysfunctional uterine bleeding. In another study by Kunz *et al* [12], prevalence of adenomyotic cyst in patients with endometriosis has been reported to be approximately 70%. In a case report by Kumar A *et al* in 2007 [13], a 32 year old woman who presented with menorrhagia and dysmenorrhea was found to have adenomyotic cyst on hysteroscopy. In the case series presented here, 77.8% of women presented with infertility and 22.2% presented with abnormal uterine bleeding.

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 just as a sub endometrial haemorrhagic area, thus get missed on these techniques. 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 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 [4] should be considered for its diagnosis including an irregular endometrium with endometrial defects, altered vascularisation, and cystic hemorrhagic lesion.

A classification system for adenomyotic cyst has been given by Brosens *et al*. [1]. He suggested the acronym MUSCLE for its classification which includes M: myometrial location (intramural, submucous, subserous), U: uterine site (midline, paramedian, lateral), S: structure (cystic, mixed, polypoid), C:

contents (clear, hemorrhagic), L: level (fundus, body, cervix), and E: (endometrial or inner lining endometrium, metaplastic). Treatment options for focal adenomyotic cysts range from medical management including use of non-steroidal anti-inflammatory drugs and hormonal suppression for symptomatic relief, to surgical resection. There are various modalities to do the surgical excision. It can be done mechanically by using 5 French scissors or via ablative procedure using energy sources.

Mechanical excision by using 5-Fr scissors during hysteroscopy allows a clear dissection of the myometrial wall of the cyst from the surrounding myometrium. Instead of dissection with scissors, an ablative technique can also be used to do the resection. The ablative procedure destroys the whole inner cystic wall, thus helping in complete excision of cyst. Also, ablative approach is preferable for those cysts which are localized deeper in the intramural portion. However, ablative procedure carries higher risk of thermal injury to endometrium and deeper structure as compared to the mechanical method of excision.

Giana M *et al*. in 2005 [14] gave a case report of 46 year old woman with intramyometrial cystic adenomyosis which was removed hysteroscopically via ablation. On transvaginal ultrasound, an anechoic area was visualised. Hysteroscopy was done which revealed a cystic mass on posterior wall of uterus. The cyst was excised by bipolar resectoscope. Histopathological examination confirmed the diagnosis of adenomyotic cyst. It was concluded that transvaginal ultrasound with hysteroscopy is specific for its diagnosis and management of adenomyotic lesion.

In 2014, S. Gordts *et al* [9] had described 2 cases of focal adenomyotic cyst, both managed hysteroscopically, one by using ablative procedure and second by mechanical excision. First case was of 51 years old woman who presented with menorrhagia. She was found to have bulging area of abnormal vascularisation on the anterior wall, which was opened with 5 Fr scissors, following which brownish fluid was drained. Histological examination confirmed the diagnosis. Another case was of 38 years old woman who presented with primary infertility since 3 years and recurrent intramural cyst. Ultrasound showed the cystic adenomyotic cyst at the isthmic level of uterus. Hysteroscopy under ultrasound guidance was done, which revealed focal adenomyotic cyst. Hysteroscopic resection was done by using spirotome and bipolar resectoscope.

In a case report by Ryo *et al*. in 2006 [15], another method for adenomyotic cyst ablation was used i.e. radio-frequency ablation. A 21-year-old woman presented with severe dysmenorrhea, supposedly caused by cystic adenomyosis. Cyst was ablated under general anaesthesia and ultrasound guidance via a radiofrequency needle which was inserted into the cyst through the cervix. The procedures took about 15 minutes and no complications were seen. This was the first case reported as adenomyotic cyst excised by radiofrequency ablation. Kindly put red coloured paragraph here.

Here, out of the nine cases, in four cases ablative method in form of Monopolar resectoscope was used while rest all cases were dealt by mechanical method with 5 Fr scissors.

CONCLUSIONS

On conclusion, adenomyotic cyst is a rare form of adenomyosis with incidence found here is 0.76%. Now-a-days 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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