

Laparoscopic Management of Adult Cystic Adenomyoma of the Uterus in Perimenopausal Woman

Swati Dubey^{1*}, Nutan Jain², Vandana Jain³, Rhythm Bhalla⁴ and Kiran Kumari Mandal⁵

¹Department of Obstetrics and Gynaecology, SMIMS, India

²Department of Obstetrics and Gynaecology, Kanpur University, India

³Department of Obstetrics and Gynaecology, Delhi University, India

⁴Department of Obstetrics and Gynaecology, Punjab University, India

⁵Department of Obstetrics and Gynaecology, PGI Chandigarh, India

*Corresponding Author: Swati Dubey, Department of Obstetrics and Gynaecology, SMIMS, India.

Received: July 07, 2020; Published: September 30, 2020

Abstract

Adenomyosis is benign gynaecological disease and is the presence of endometrial glands and stroma in the myometrium with hyperplasia of smooth muscle. Adult cystic adenomyoma is a cystic lesion in the myometrium, a rare disease and presents as intense dysmenorrhoea and chronic pelvic pain. We report a case of a 40 year old woman presented with severe dysmenorrhoea and menorrhagia which worsened over past 3 years. Transvaginal ultrasonography revealed a globular uterus with a cystic adenomyoma of approximately 79 x 62 mm and excised successfully laparoscopically. The mass had no communication with the endometrium and both ovaries appeared normal. The findings were later confirmed by histopathological examination of surgically enucleated mass.

Keywords: Laparoscopic; Adult Cystic; Adenomyoma

Abbreviations

IUI: Intrauterine insemination; IVF: *In-Vitro* Fertilization; LDH: Lactate Dehydrogenase; ART: Artificial Reproductive Technique; ACUM: Accessory and Cavitated Uterine Mass; TVS: Transvaginal Sonography

Introduction

Adenomyosis is benign gynaecological disease with presence of endometrial glands and stroma in the myometrium with hyperplasia of smooth muscle causing globular enlargement of uterus. It usually occurs in parous women and sometimes is indication for hysterectomy. 20% of post-hysterectomy uterine specimens have diffuse adenomyosis [1]. Etiology is unknown. There are 2 varieties of this disease - diffuse or cystic. Cystic adenomyoma is of two types - juvenile or adult type. Juvenile cystic adenomyoma presents as severe dysmenorrhoea, menorrhagia. Adult cystic adenomyoma presents as intense dysmenorrhoea, chronic pelvic pain and infertility.

Occasionally, a cystic lesion develop in myometrium of adenomyotic uterus, cyst is usually < 5 mm in diameter. Large cystic lesion > 1 cm in diameter is termed as adenomyotic cyst or cystic adenomyoma [2]. We present a case report of adult cystic adenomyoma in a uterus with diffuse adenomyosis. This cystic adenomyoma is diagnosed on transvaginal ultrasonography before surgery and excised successfully laparoscopically.

Case Report

A 40 year old nulliparous woman presented to us with complains of infertility, severe dysmenorrhoea and menorrhagia, worsening over last 3 years. Her menarche was at 13 years of age and was asymptomatic until few years back. She developed worsening dysmenorrhoea with excessive flow and passage of clots. Her fertility treatment included 3 cycles of IUI and 1 cycle of IVF done in 2017. Her previous reports of ultrasonography and magnetic resonance imaging examination showed well defined oval altered signal intensity lesion of size 65 x 75 x 63 mm, diagnosed as submucosal fibroid with cystic degeneration. Her LDH was 253 and CA 125 value was 451. Her general physical examination was within normal limits. On abdominal examination, 20 weeks mass felt. On bimanual pelvic examination, uterus not felt separate from the mass. Her transvaginal scan done at our hospital revealed a globular uterus with a cystic lesion of approximately 89 x 72mm with low level homogenous echoes almost looking like an endometrioma. The mass had no communication with the endometrium.

With these clinical findings, patient taken up for laparoscopy and hysteroscopy. Pneumoperitoneum achieved by using left lateral non umbilical point (Jain point) followed by 5 mm trocar insertion [3,4]. As Jain point is ergonomically compatible for large masses also, it was preferred point of entry in our case. Whole abdominal cavity completely visualized by using 5 mm telescope and 10 mm port site optimized. 3 more accessory working ports made. Intra operatively, a big posterior wall bulge noted. Diluted vasopressin (20 units in 300 ml of normal saline) injected into the uterus over the bulge. A vertical incision given on the most bulging part of the mass with the Harmonic ACE. Profuse thick chocolate coloured content escaped out of cavity and pre operative diagnosis of cystic adenomyoma confirmed. Cyst lining was then carefully enucleated from the surrounding myometrium. The uterine cavity not entered. Suturing done in three layers in a continuous running manner for better approximation of the layers.

Surgery had minimal blood loss. Her intra and post-operative period was uneventful. Patient discharged next day. Her husband's semen analysis was normal. She was advised to undergo IVF/ART after 3 months.

Histopathology examination revealed a cystic structure lined by endometrial epithelium and surrounded by myometrial tissue, consistent with cystic adenomyoma.

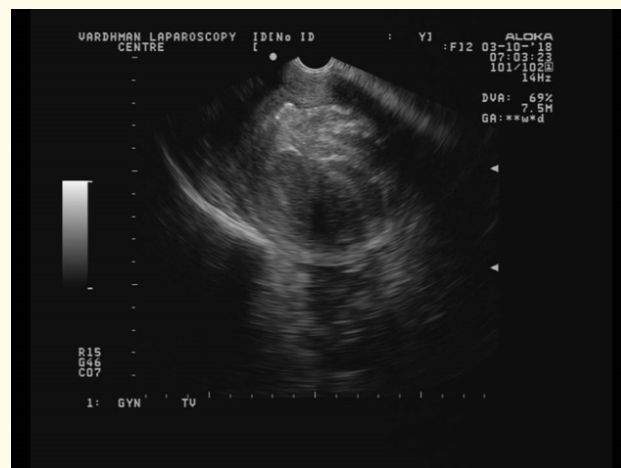


Figure 1: Ultrasound image of cystic adenomyoma.

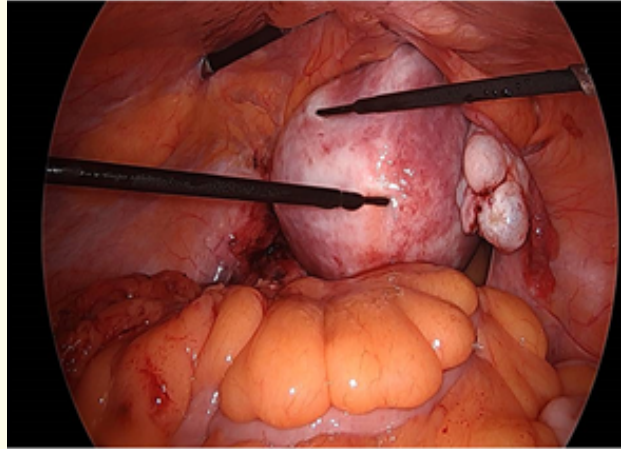


Figure 2: Pitressin injected.

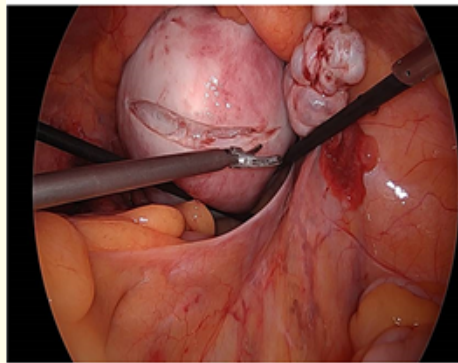


Figure 3: Incision with harmonic ACE.

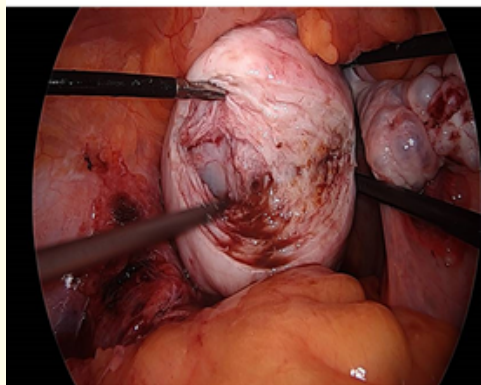


Figure 4: Chocolate color fluid drained out of cystic adenomyoma

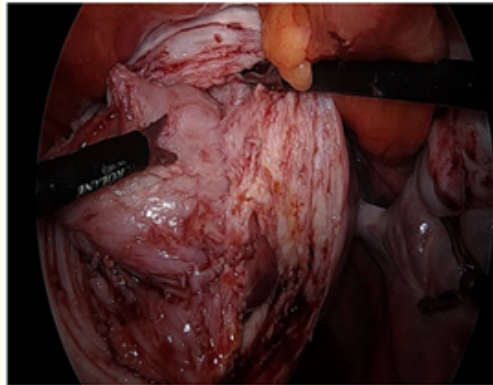


Figure 5: Enucleating cystic adenomyoma.

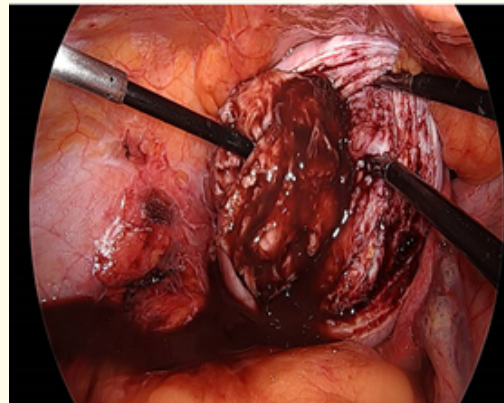


Figure 6: Cystic adenomyoma with chocolate coloured fluid.

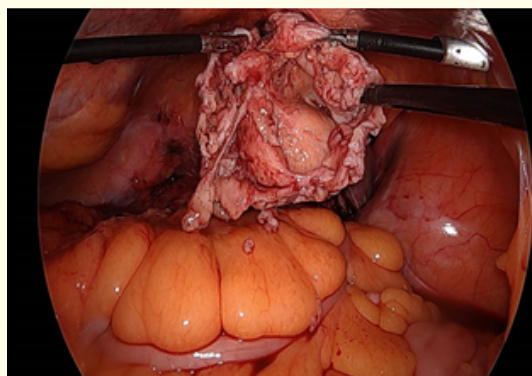


Figure 7: Cystic adenomyoma.

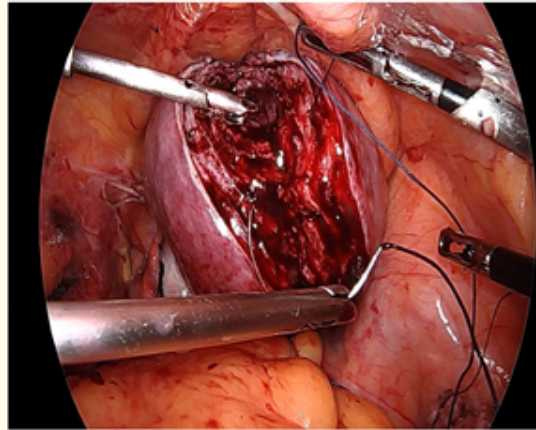


Figure 8: Endosuturing of the adenomyoma bed in layers to obliterate dead space.

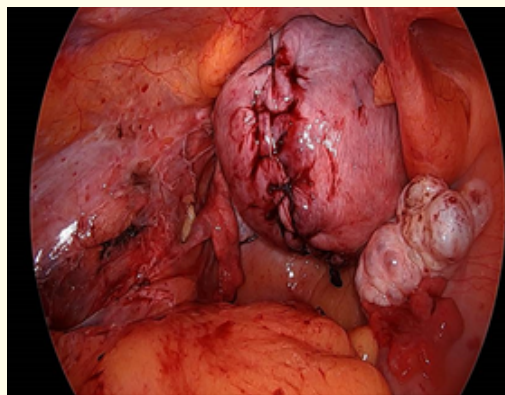


Figure 9: End laparoscopic view

Discussion and Conclusion

Cystic adenomyoma is a rare disease resulting from extensive haemorrhage within ectopic glands in the myometrium. It is characterized by progressively intractable dysmenorrhoea. It is similar to eutopic endometrium during menstruation, when a blood filled cavity similar to endometrioma presses against the surrounding myometrium. Cyst contains chocolate like fluid [5]. Juvenile cystic adenomyoma is the result of uterine anomaly found in younger age group (≤ 18 years) [1]. In contrast adult cystic adenomyoma is rare adenomyotic condition¹ found in multiparous woman > 30 years of age.

Early occurrence of the symptoms soon after menarche shows that juvenile cystic adenomyoma is a congenital malformation caused by defect in development of mullerian ducts. Most common location of this lesion is anterior uterine wall at the level of round ligament insertion [6]. Accessory uterine mass could be caused by duplication or persistence of ductal mullerian tissue at the insertion of round ligament, possibly due to a gubernaculum dysfunction. In our case, it is located at posterior surface of uterus.

Whereas adult type is result of parturition or following C-section, myomectomy and transcervical curettage [7]. As per cases reported, pre operative diagnosis can be confused with leiomyoma with cystic degeneration, cystic ovarian tumour, congenital uterine cyst, like in our case, which can be confirmed by surgical exploration only. For accurate diagnosis and management this diagnosis should be kept in mind.

Acien, *et al.* [8] proposed the term accessory and cavitated uterine mass (ACUM) with functional endometrium. Its inclusion criteria is : (1) isolated accessory cavitated mass usually located under round ligament; (2) normal uterus, tubes, ovaries; (3) surgical case with excised mass, histopathological examination; (4) accessory cavity lined by endometrial epithelium; (5) chocolate coloured fluid contents; (6) no adenomyosis in uterus, unlike our case which had diffuse adenomyosis. In our case, it was not in relation to round ligament and was posteriorly placed. Ultrasound image strongly suggested diffuse adenomyosis which was further reconfirmed during laparoscopy.

This clinical entity presents diagnostic difficulties and can be missed, overlooked and at times wrongly diagnosed or simply not kept under consideration. Like in our case, MRI misdiagnosed this lesion for fibroid with cystic degeneration. While investigating cases with pelvic pain, one should learn the art of diagnosing it on TVS as priority in imaging modality. 3D ultrasound has been proposed as an accurate diagnostic aid in determining myometrial border of adenomyosis [9].

Surgical enucleation is gold standard treatment of these benign lesions. Adenomyotic cyst is better delineated than adenomyosis from surrounding myometrium but lesser than uterine myomas. In our case, laparoscopic enucleation was performed as fertility enhancing surgery.

It is compelling to create awareness of this entity which is repeatedly misdiagnosed as myoma. Juvenile cystic adenomyoma is commonly reported but our patient presented as case of adult cystic adenomyoma with complaints of severe dysmenorrhoea and menorrhagia and hence, rightly said perimenopause is not the end.

Conflict of Interest

No conflict of interest.

Bibliography

1. Chun SS, *et al.* "Juvenile cystic adenomyoma in a 19-year-old woman: a case report with a proposal for new diagnostic criteria". *Journal of Laparoendoscopic and Advanced Surgical Techniques A* 21 (2011): 771-774.
2. Takeda A, *et al.* "Laparoscopic management of juvenile cystic adenomyoma of the uterus: report of two cases and review of the literature". *Journal of Minimally Invasive Gynecology* 14 (2007): 370-374.
3. Jain N, *et al.* "Jain point: A new safe portal for laparoscopic entry in previous surgery cases". *Journal of Human Reproductive Sciences* 9.1 (2016): 9-17.
4. Sharp H. "Overview of gynecologic laparoscopic surgery and non-umbilical entry sites". Uptodate (2019).
5. Calagna G, *et al.* "Cystic adenomyosis spreading into subserosal-peduncolated myoma: How to explain it". *International Journal of Surgery Case Reports* 8 (2015): 29-31.
6. Acien P, *et al.* "The cavitated accessory uterine mass: A müllerian anomaly in women with an otherwise normal uterus". *Obstetrics and Gynecology* 116 (2010): 1101-1109.
7. Wang JH, *et al.* "Single large cystic adenomyoma of the uterus after cornual pregnancy and curettage". *Fertility and Sterility* 88 (2007): 965-967.

8. Acien P, *et al.* "New cases of accessory and cavitated uterine masses(ACUM): a significant cause of severe dysmenorrhoea and recurrent pelvic pain in young women". *Human Reproduction* 27 (2012): 683-694.
9. Koukoura O, *et al.* "Laparoscopic treatment of a large uterine cystic adenomyosis in a young patient". *BMJ Case Reports* (2015).

Volume 9 Issue 10 October 2020

©All rights reserved by Swati Dubey, *et al.*