

Laparoscopic Management of Juvenile Cystic Adenomyoma

Dr C.P. Dadhich; Dr Nidhi Mehta; Dr . Tripti Dadhich

Corresponding Author: Dr.Nidhi Mehta

Abstract:

Background: Juvenile cystic adenoma is an uncommon type of adenomyosis . It affects young girls and usually manifests as severe dysmenorrhea and recurrent pelvic pain. Aim of study is to raise awareness about ACUM ;its characterization;location;early diagnosis and appropriate treatment.

Materials And Methods: Laparoscopic excision of cystic adenomyoma of the uterus occurring in 5 young women is reported which were misdiagnosed pre-operatively as a hematometra in a non-communicating horn of the unicornuate uterus.

Results: Histopathologic examination of the resected tissues showed the presence of an endometrial structure composed of epithelium and stroma within myometrial nodule. In these patients, dysmenorrhea disappeared postoperatively.

Conclusion: Surgery in form of operative laparoscopy is helpful. Early diagnosis and excision will completely eliminate the cause of pain.

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I. Introduction

Juvenile cystic adenomyoma (JCA) OR ACUM (Accessory Cavitated Uterine Masses) is a rare form of adenomyosis. It affects young girls and usually manifests as severe dysmenorrhea and recurrent pelvic pain. Patients are often misdiagnosed to have mullerian anomalies such as a unicornuate uterus with hematometra in a non-communicating rudimentary horn or a bicornuate uterus with segmental atresia because of the typical clinical presentation and the rarity of the condition.(1). In 2009, Hiroyuki *et al*¹ defined the **diagnostic criteria of juvenile cystic adenomyoma** :

1. age (<30 years)
2. presence of cystic lesion ≥ 1 cm in diameter independent of uterine lumen
3. covered by hypertrophic myometrium on diagnostic images
4. associated with severe dysmenorrhea.

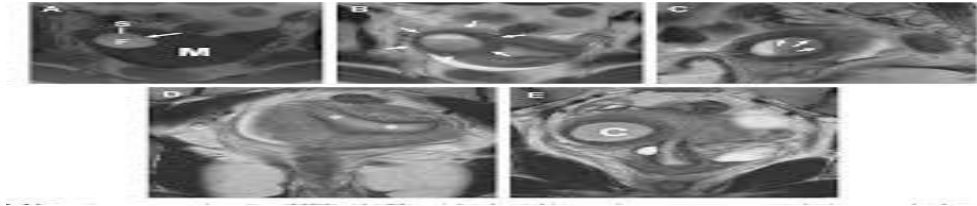
AIMS AND OBJECTIVES

1. To raise the awareness about ACUM (accessory cavitated uterine masses) as a different entity from adult adenomyosis.(3)
2. Determining the characterization and localization of the lesion
3. To review the methods for correct diagnosis of ACUM or juvenile cystic adenomyoma.
4. Early diagnosis and laparoscopic excision of the lesion remains mainstay of treatment.

II. Material And Methods

5 Patients under the age group of 13-20 years presented to us with complaints of severe dysmenorrhea and recurrent pelvic pain which was non-responsive to medical management. They had regular menstrual cycles but with progressive dysmenorrhea throughout the cycle. On sonographic evaluation, mass with central cystic degeneration was seen along the right lateral aspect of the uterus and medial to the right ovary. The differential diagnosis given were hematometra in the non-communicating rudimentary horn of unicornuate uterus, endometriosis, degeneration of myoma and torsion of ovarian mass. MRI findings were suggestive of uterine bicornis with right horn hematometra and in 1 case MRI was suggestive of broad ligament fibroid. Complete resection of the cystic adenomyoma was performed laparoscopically in all patients. The lesion was unencapsulated (unlike myoma) and locally defined to the uterine myometrium (unlike diffuse adenomyosis), and there was chocolate-colored blood in the cavity. No other müllerian anomaly was observed in any patient.

MRI SHOWING CYSTIC ADENOMYOMA



III. Results

Histopathologic analysis revealed features suggestive of adenomyosis in all cases. After surgery, dysmenorrhea resolved completely in all patients. Compared with preoperative visual analog scores, significant improvement was observed at the first menstrual cycle after surgery. Patients were followed up every 3 months for a minimum of 12 months to detect development of dysmenorrhea or any other menstrual disorder.



FIGURE 1 CYSTIC MASS SEEN JUST ABOVE ROUND LIGAMENT

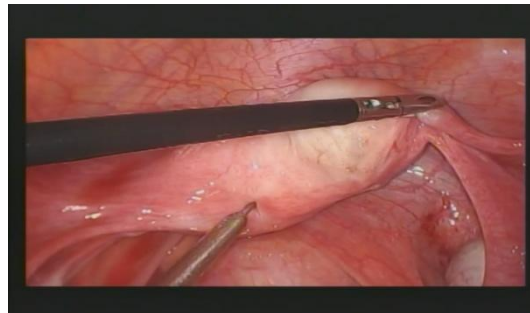


FIGURE2 VASOPRESSIN BEING INJECTED INTO THE MASS



FIGURE 3 INCISION OVER THE MASS



FIGURE 4 CHOCOLATE CLOURED FLUID BEING DRAINED

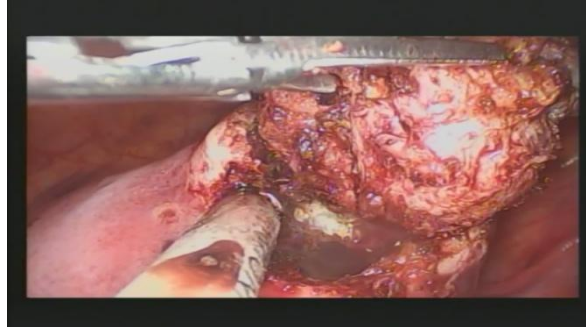


FIGURE 5 COMPLETE EXCISION OF ADENOMYOMA



FIGURE 6 AFTER REMOVAL OF MASS

IV. Discussion

Uterine adenomyosis is a benign proliferative disease of unknown etiology in which the myometrium is invaded by endometrial tissue composed of endometrial glands and stroma, and is most frequently found in multiparous women over 30 years old. Juvenile cystic adenoma is an uncommon type of adenomyosis. The pathognomonic clinical feature of juvenile cystic adenomyoma is its early onset of severe dysmenorrhea that usually starts with menarche. This symptom could be attributed to intracystic bleeding and stretching of the cystic cavity. (4) Medical treatment with GnRH agonist only provide temporary relief, symptoms resumed with the return of menstruation. Recently, these adenomyomas are categorized under accessory and cavitated uterine masses. its peculiar location is always on the anterior wall of the uterus near the origin of the round ligament. This may be explained by the duplication of mullerian tissue at the level of the insertion of the round ligament which in turn maybe because of a defect in the female gubernaculum. MRI can be used for a non-invasive diagnosis. The gold standard treatment for a juvenile cystic adenomyoma is complete excision of the lesion, and use of minimally invasive method has been described.(5)

V. Conclusion

Juvenile cystic adenomyoma or ACUM(Accessory Cavitated Uterine Masses) could be a new variant of adenomyoma that requires early surgical intervention. Operative laparoscopy in the hands of an experienced surgeon is helpful. Early surgical treatment should be considered the treatment of choice as it will completely eliminate the cause of pain.(6)

References

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VI. Review of Literature

Study	Diagnosis	Patient age at diagnosis (yrs)	Signs/symptoms	Age at initial onset of signs/symptoms (yrs)		Diagnostic procedures	Location of nodule	Size of cystic cavity (cm)		Surgical treatment procedure	Previous treatment	Histologic evidence of adenomyoma tissue
				menarche (yrs)	signs/symptoms (yrs)			value (normal < 35 U/mL)	Serum CA 125			
Tamura et al ⁷	Juvenile adenomyotic cyst of the corpus uteri	16	Severe menorrhagia	12	15	US, MRI, DIP	Left/posterior	3	N/A	Diagnosis under laparoscopy and resection by laparotomy	N/A	Yes
Nabeshima et al ⁸	Cystic adenomyoma	19	Severe dysmenorrhea	N/A	N/A	US, MRI, HSG, intraoperative hydroultrasoundography	Right/posterior	3	40.9	Total laparoscopic resection	GnRH agonist	Yes
Takeda et al (current report)	Juvenile cystic adenomyoma	20	Severe dysmenorrhea	13	13	US, MRI, HSG, DIP	Right/anterior	1.5	25	Total laparoscopic resection	NSAIDs, intermittent administration of GnRH agonist since 17 years old	Yes
Takeda et al (current report)	Juvenile cystic adenomyoma	20	Severe dysmenorrhea	14	14	US, MRI, HSG, DIP, intraoperative 3-D US	Left/anterior	1.1	40.5	Laparoscopic-assisted resection	NSAIDs	Yes

3-D US = 3-dimensional ultrasonography; DIP = drip infusion pyelography; GnRH = gonadotropin-releasing hormone; HSG = hysterosalpingogram; MRI = magnetic resonance imaging; N/A = not available; NSAIDs = nonsteroidal anti-inflammatory drugs; US = ultrasonography.