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Case Report

Accessory cavitated uterine mass: A rare cause of severe dysmenorrhoea managed by minimally invasive surgery

Cyriac Pappachan[1], Manasa Saraswathy Kabalimurthy[1]*, Roshni Subhash[1], Koothan U T[1]

[1]*Dept. of Gynaecological Endoscopy, Lifeline Superspeciality Hospital, Adoor, Kerala, India*

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ABSTRACT

Accessory cavitated uterine mass (ACUM) is a rare congenital mullerian anomaly, encountered in young, nulliparous women. It is a non-communicating cavity, lined by functional endometrium in an otherwise normal uterus, presenting with severe intractable dysmenorrhoea. On ultrasound, it is a striking myometrial lesion but often misdiagnosed due to unfamiliarity. Though studies have described medical management, recurrence of symptoms occurs after discontinuing treatment. Surgical management, either conservative, by excising the lesion completely, or hysterectomy, can resolve the symptoms. Here, we report a 29-year old nulligravida with primary infertility presenting with severe dysmenorrhoea, unresponsive to analgesics. She was preoperatively diagnosed to have an ACUM and was managed by conservative minimally invasive surgery. Histopathology showed endometrium with secretory activity surrounded by myometrium, consistent with our findings. On postoperative follow-up, patient reported complete recovery from dysmenorrhoea. Appropriate diagnosis of this rare entity is a true challenge because of lack of awareness and the wide range of close differential diagnoses. It is imperative to understand and distinguish this peculiar lesion, with an ultimate aim of performing timely surgery, for complete symptom alleviation, especially when fertility is a concern.

1. Introduction

Accessory cavitated uterine mass (ACUM) is a rare congenital mullerian anomaly. It is a non-communicating cavity, lined by functional endometrium in an otherwise normal uterus.[1] This cavitated lesion is located in the lateral aspect of myometrium, inferior to the attachment of round ligament. This entity encountered in young, nulliparous women, poses a significant cause of severe intractable dysmenorrhoea, and chronic pelvic pain, usually unresponsive to analgesics.[2–6] Some women also present with subfertility.[7] Ultrasound, MRI and hysteroscopy serve as useful diagnostic tools. Even with these facilities for detailed visualisation and examination of the uterus, ACUM is still frequently misdiagnosed. Appropriate diagnosis of this rare entity is a true challenge because of lack of awareness about this lesion and the wide range of close differential diagnoses. Here, we report a 29-year old nulligravida with primary infertility who presented with severe dysmenorrhoea. She was preoperatively diagnosed to have an ACUM and was managed by conservative minimally invasive surgery.

1.1. Informed consent

All procedures followed the ethical standards of the responsible committee on human experimentation (institutional and national) and the 1975 Declaration

* *Corresponding author*.
E-mail address: manasacdm@gmail.com (M. S. Kabalimurthy).

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of Helsinki, and as revised in 2000. Informed consent was obtained from the patient. IRB approval was exempt as this is a case report involving a single patient.

2. Case Report

A 29-year old nulligravida, married for 3 years, presented with a 2-year history of severe dysmenorrhoea. She attained menarche at 14 years. She had regular cycles and normal flow. Patient was referred to us with an ultrasound report suggestive of left ovarian cyst and an MRI report of a bicornuate uterus. MRI performed in our centre showed an anteverted normal sized uterus with normal endometrium. There was a well circumscribed spherical mass with echogenic fluid content, surrounded by a layer of normal myometrium. The lesion was located in the left posterolateral myometrium, measuring ~ 2x2cm. Bilateral ovaries appeared normal. With a provisional diagnosis of ACUM, a decision to proceed with laparoscopic excision of the lesion was made. Hysteroscopy was performed, which showed a normal uterine cavity, and normal bilateral ostia, ruling out the possibility of a bicornuate uterus, reinforcing our diagnosis. Laparoscopy revealed a uterus with a 2x2cm projection in the left posterolateral wall of the uterus, below the round ligament, mimicking a subserous fibroid, but cystic in consistency. Superficial endometriotic lesions were present at the attachment of uterosacral ligaments on uterus. Both ovaries were normal. Chromopertubation was performed, revealed patency of both fallopian tubes. Dilute vasopressin (1 : 20 dilution) was injected onto to adjacent myometrium. The ACUM lesion was enucleated in toto and excised completely with harmonic scalpel, without entering the uterine cavity. Chocolate coloured material was drained during excision of the lesion. The incision was sutured in layers with number 1 barbed sutures. Superficial endometriotic lesions on the posterior surface of uterus and uterosacral ligaments were fulgurated. Specimen retrieval was done in an endo-bag. Patient had an uneventful post operative recovery and was discharged the next day. Upon postoperative follow up, she reported complete recovery from dysmenorrhoea, and ultrasound showed no abnormality. She has been advised to plan for pregnancy after 3 months. Histopathological examination confirmed a cystic mass lined by endometrium with secretory activity surrounded by myometrium, consistent with our findings. The histopathological report along with the operative findings confirmed the diagnosis of ACUM.

3. Discussion

There are case reports suggestive of ACUM, using various terms like ‘juvenile cystic adenomyoma’,[5,6] ‘isolated cystic adenomyoma’.[4] The term ‘ACUM’ was coined by Acién et al. in 2012, to describe these lesions, which until then had no agreed name.[1] Poor recognition of this abnormality

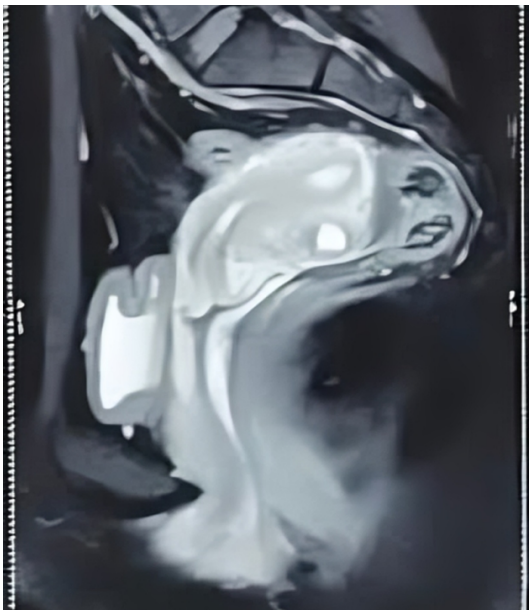

Figure 1: MRI showing accessory cavitated uterine mass

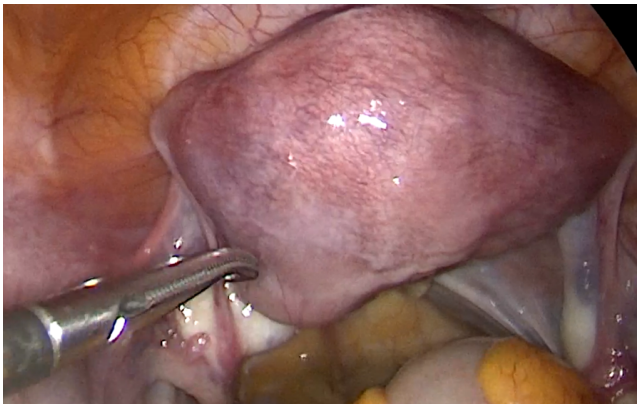

Figure 2: Laparoscopic view: Projection in the posterior aspect of uterus

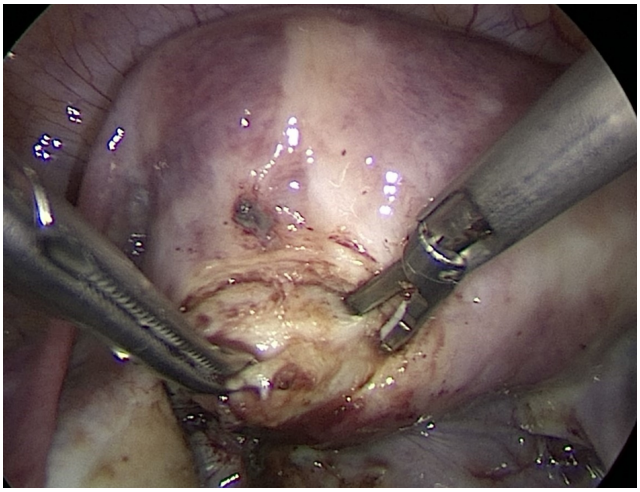

Figure 3: Incision made over the lesion with the Harmonic scalpel

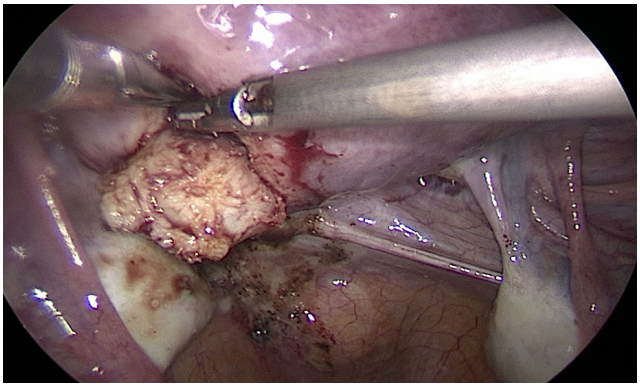

Figure 4: Enucleation of the ACUM with the Harmonic scalpel

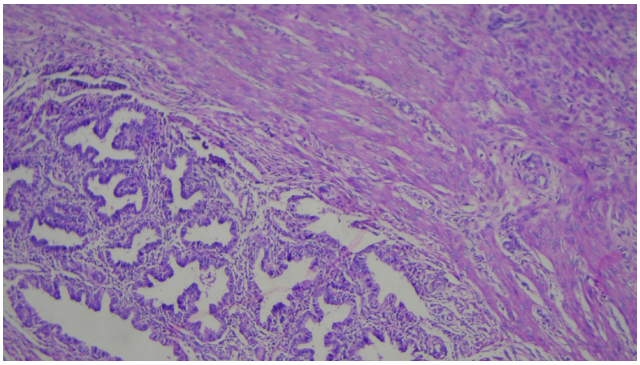

Figure 5: Histopathology showing endometrium with secretory activity surrounded by myometrium, consistent with our findings

by gynaecologists and radiologists, explains the paucity of literature evidence, which is limited to few case reports and case series. The true prevalence of ACUM, is therefore unknown.

Acien et al. have suggested two theories about the development of ACUMs -an isolated malformation of the mullerian duct or a dysfunction of female gubernaculum.[1] It is well-known that the remnants of wolffian duct can occasionally give rise to Gartner cyst. Similarly, congenital uterine anomalies can arise from the remnants of mullerian ducts. Duplication and persistence of a mullerian duct segment at the level of attachment of round ligament can be associated with the origin of ACUM. The round ligament of the uterus is the embryological remnant of the female gubernaculum. Acien et al hypothesise that the normal development of uterus is induced by the attachment of gubernaculum to mullerian ducts. The fact that all cases of ACUM describe the location near the insertion of round ligament or its pathway, reinforces the theory that ACUM develop from dysfunction of female gubernaculum.[1]

The most common presenting complaint in women with ACUM is severe dysmenorrhoea, followed by chronic pelvic pain. Some women also have infertility and dyspareunia. Severe pain can be attributed to the accumulation of menstrual fluid shed from a functional endometrium inside an enclosed space, leading to stretching of the cavity.

Since ultrasound is the first line tool of imaging in gynaecological clinics, awareness about the ultrasound features of ACUM is imperative to aid better detection in everyday practice. ACUM appear as cavitated, spherical lesion containing echogenic fluid, surrounded by normal myometrium, located typically inferior to the presumed insertion of round ligament. If ultrasound is inconclusive, MRI can facilitate the diagnosis of ACUM.[8]

The present case reported, was also misdiagnosed as an ovarian cyst and a bicornuate uterus in another centre. The presence of a morphologically normal uterine cavity and visualisation of two normal interstitial portions of fallopian tubes rules out obstructive congenital uterine malformations. ACUM can be confused with an endometrioma adherent to uterus, but the presence of characteristic myometrial mantle and absence of surrounding follicles in the ovarian cortex clinches the diagnosis of ACUM. True cystic adenomyomas tend to affect older, parous women, who present with heavy painful menses, with other features suggestive of adenomyosis on ultrasound examination. The internal endometrial lining is typically absent in true cystic adenomyomas and is surrounded by disorganised myometrial tissue.[3]

Since the predominant symptom of ACUM is pain, NSAIDs can be prescribed, but most case reports have declared its inadequacy for pain control. Continuous oral contraceptive pills, GnRH injections, and levonorgestrel intrauterine system have achieved pain relief but many patients perceived recurrence of symptoms after stoppage of treatment.[1,2,4–6]

The ultimate and definitive treatment for ACUM, is surgery. All reported cases of ACUM that have undergone surgical management have reported complete symptom relief till completion of follow up.[1,2,9]

Surgical management has been by excision of ACUM or even by hysterectomy in selected women with separate indications.[10] Route of surgery can be by laparotomy or laparoscopy. We preferred to operate by laparoscopy because of the obvious advantages of quicker recovery, reduced postoperative pain and also the precise enucleation of lesion, which can be achieved owing to magnification and fine dissection instruments.

4. Conclusion

ACUM, is an underdiagnosed treatable cause of severe progressive dysmenorrhoea in young females. A high index of suspicion is needed to diagnose the lesion. Appropriate diagnosis of this rare entity is a true challenge because of lack of awareness and the wide range of close differential diagnoses. It is imperative to understand and distinguish this peculiar lesion, with an ultimate aim of performing timely

surgery, for complete symptom alleviation, especially when fertility is a concern. Laparoscopic excision is the appropriate, minimally invasive option for the management of ACUM. Further large-scale studies, evaluating the effect of ACUM excision on fertility holds an interesting research arena in the future.

5. Sources of Funding

None.

6. Conflict of Interest

None.

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Author biography

Cyriac Pappachan, HOD

Manasa Saraswathy Kabalimurthy, Fellow in Gynaecological Endoscopy https://orcid.org/0009-0003-2355-5201

Roshni Subhash, Consultant

Koothan U T, Consultant

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