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CASE REPORT

Laparoscopic Approach to Accessory and Cavitatory Uterine Mass(ACUM): A Report of Four Patients in a Year

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Introduction

This is a rare form of Mullerian anomaly which presents in women with chronic unilateral recurrent episodes of dysmenorrhoea and non-cyclical abdominal pain [1]. It is an accessory mass at the insertion of round ligament that is usually lined by normal functioning endometrium. This requires expertise in diagnosis as it is rare and is one of the treatable cause of severe dysmenorrhoea. There is a difficulty in diagnosis of ACUM because of its rarity and similar clinical presentation which mimics rudimentary and cavitated uterine horns as those found in other uterine malformations such as bicornuate uterus, adenomyosis with cystic degeneration, degenerated leiomyomas. To our knowledge, about less than 60 cases have been reported so far and in this we present a case series of four patients diagnosed as ACUM at various ages of presentation.

Case presentation

Case 1

This 27-year-old Para 2 Live 2, 2 caesarean sections, presented with complaints of chronic right sided dysmenorhoes with pain radiating to the right leg for more than a decade. She has consulted various other specialities like orthopedic surgeon, nuerosurgeon for the same and currently she was resistant to all the pain killers. She gives a history of bicornuate uterus which was diagnosed during her first delivery. In suspicion for ACUM, we requested an MRI, which showed a heterogenous lesion in right lateral wall of uterus, measuring $4.1 \times 2.5 \times 2.7$ cm, inferior to right cornua. Endometrial cavity of this lesion measures approximately 9 mm and shows fluid fluid level in T2W1 which is hyperintense in T1W1 representing hemorrhage. No demonstrable communication between this endometrial cavity with the normal endometrial cavity was noted. We performed a Laparoscopic excision of the Acessory uterine mass along with diagnostic hysteroscopy. We did en bloc excision and diagnostic hysteroscopy revealed no communication of the accessory cavity to the original endometrial cavity.

Case 2

A 39 years, married for 16 years, nulliparous presented with complaints of chronic left sided dysmenorrhoea radiating to left leg for the past 15 years. She has consulted at various places for which ultrasound was done which showed intramural fibroid with cystic degeneration measuring 3.3×2.7 cm in the anterior myometrium near the fundus. Over the years, the ultrasound showed only the persistent intramural fibroid with cystic degeneration. We had suspicion of ACUM as this intramural fibroid was unlikely the cause of dysmenorrhoea. We requested for an MRI which confirmed the diagnosis of ACUM. We did a Laparoscopic excision with diagnostic hysteroscopy and excised the mass (Figs. 1 and 2).

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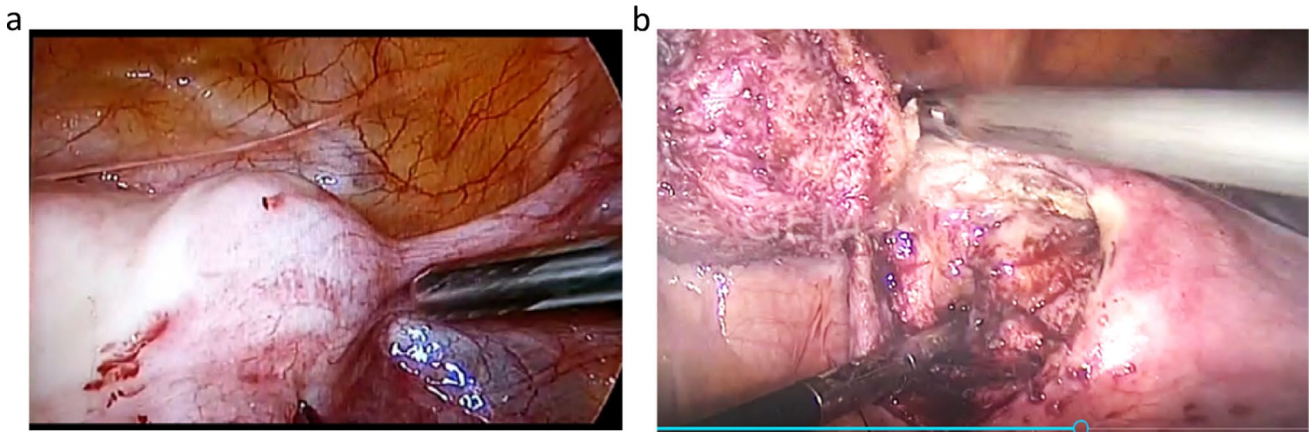

Fig. 1 **a** Laparoscopic view of ACUM near the insertion of the round ligament. **b** Intraoperative picture while enucleation of the mass

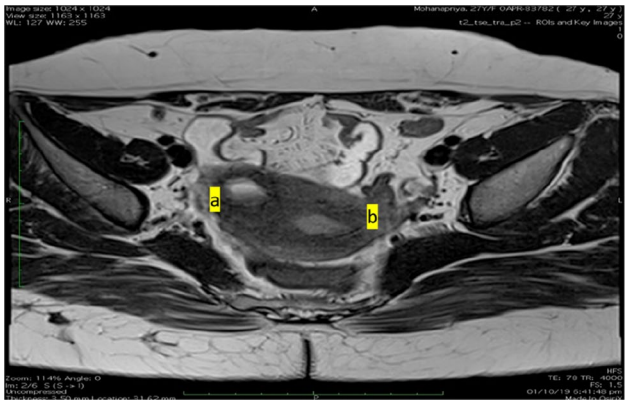

Fig. 2 Pre operative MRI showing two endometrial cavities

Case 3

A 22-year-old unmarried lady presented with complaints of progressively increasing dysmenorrhoea since menarche. She was evaluated in many places following which ultrasound was done which was suggestive of right lateral wall fibroid with cystic degeneration. MRI was suggested here which showed a well-defined homogenous lesion measuring 2.5 × 2.3 × 2.4 cm with surrounding thick hypo intense margin in the right lateral wall of the fundus. The lesion is seen intending the right cornua of the endometrial cavity- s/o accessory cavitary uterine mass. Intra operatively, Uterine contour appears normal externally except a bulge on the right lateral aspect of fundus just below the insertion of round ligament. We excised the mass laparoscopically.

Case 4

A 15-year-old unmarried girl presented with severe dysmenorrhoea since menarche with increasing non-cyclical abdominal pain since 2 years. Ultrasound was done elsewhere commented that there is an endometrial like fluid collection within the uterus at the right cornua. Since the definitive diagnosis could not be made she was referred here for further management. An MRI was done here which showed a bulky uterus with a 3 × 2.8 cm intramural lesion in the right lateral wall—Benign endometrioma. From her clinical symptoms and MRI were discussed and diagnosis of ACUM was confirmed. She underwent laparoscopic excision of the accessory mass.

Technique

Under General anaesthesia, the patient is in supine position, veress entry was followed for pneumoperitoneum. Supraumbilical 10 mm camera port was used and two ipsilateral right lateral port each 5 mm was made and one left lateral 5 mm assisting port was made. In all our four cases presented in a spam of one year, the mass presented as a bulge near the insertion of the round ligament, where in there was asymmetrical enlargement of the uterus, Bilateral fallopian tubes and ovaries were normal, no e/o endometritic deposits or fibrosis were noted in all the four cases. Among the four cases, three cases ACUM was localised to the right side and one case it was localised to the left side. About 100 ml of diluted vasopressin (about 3 U in 100 ml) was instilled over the mass. Myoma screw was used for manipulation. We prefer to use a transverse incision over the mass, and the mass was dissected from the uterus without breeching the main endometrial cavity.

The cleavage plane was not easy during dissection as certain amount of adenomatous changes was also expected during the dissection. The mass was excised in to without entering the main endometrial cavity as all the masses were localised to one of the sides near the insertion of the round ligament. During the dissection, out of the 4 masses one mass emitted chocolate coloured fluid and was thoroughly suctioned out. The myometrial defect was closed in two layers of barbed sutures (1–0 V loc). The specimen was placed in the endobag, spliced within the bag, where in we were able to get the closer view of the endometrial cavity. All the specimen with the endobag was taken out through the supra umbilical 10 mm port without any spillage. A thorough irrigation and suctioning was done. An adhesion barrier like interceed was placed over the wound.

The entire specimen was sent for histopathological evaluation which revealed myometrial tissue with hemosiderin laden macrophages with the endometrial lining and occasional endometrial glands.

Discussion

Mullerian anomalies accounts for 4–7% of the population and they may lead to significant reproductive outcomes. Unlike the other anomalies these uterine like masses are a distinct entity which are composed of hormone sensitive smooth muscle cells with functional endometrium arranged in the periphery of a normal appearing uterus. The pathogenesis may be related to ductal duplication of the gubernaculum dysfunction at the level of attachment of the round ligament [2]. Hence they have to be considered as a separate entity rather than classifying them as a uterine anomaly. However, according to Pooya et al. [1], the criteria for diagnosis depend on normalcy of the uterus, tubes and ovaries, isolated accessory cavitary with functioning endometrium confirmed intraoperatively by the appearance of chocolate coloured material (collected endometrial blood) and pathologically by the presence of functioning endometrium.

These patients were followed up after three to nine months following surgery. We noticed no recurrence of symptoms up to date and the patients are symptoms free from surgery. According to Majumdar et al. [3], chromopertubation was done to see the patency of the tubes and also to see that no direct communication of the main uterine cavity to the mass was noted. However, we have done diagnostic hysteroscopy in two of our patients to find out the direct communication of the endometrial cavity. The surgeon should always focus on en bloc surgical resection[2] of the remaining adenomyotic lesion in the myometrium to ensure good clearance as there is no definitive cleavage planes as we notice in myomectomy. Also the radiological diagnosis is also very important to make the conclusion and the radiologists should be clearly told about this rare entity before making the diagnosis as earlier these were referred to as isolated adenomyotic cyst [4]. There are a few MRI features [5], according to which the ACUM is lateralised either to right or to the left side of the uterus with regular borders located beneath the insertion of round ligaments with central hypointense on T1 and the cavity is surrounded by low T1 and T2 signal corresponding to the normal junctional zone.

Conclusion

ACUM is a treatable cause of dysmenorrhoea if a good history taking and early diagnosis is made. 3 D ultrasonography and MRI aid in its diagnosis. Early intervention with laparoscopy or with laparotomy ameliorates the symptoms and have good clinical outcomes. Persistent chronic dysmenorrhoea and in the imaging mostly looks like an isolated type 5 or type 6 leiomyoma with the functioning endometrium with normal appearing uterus and adnexa should always rise the clinical suspicion of ACUM.

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Declarations

Conflict of Interest The authors share no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards". Informed consent was obtained from all individual participants included in the study.

Human and Animal rights There is no violation of human rights.

Informed Consent Informed written consent for publication of this article was obtained from patients\ and her relative.

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