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Case Report

Two case reports of accessory cavitated uterine mass-diagnostic challenges

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ABSTRACT

Accessory and cavitated uterine mass (ACUM) is a rare Mullerian duct anomaly of unknown incidence, affects young women. ACUM symptoms such as dysmenorrhea and chronic pelvic pain (CPP) resistant to common analgesics and hormonal contraceptives. Here we report two ACUM cases in our hospital within one year of period with different clinical manifestations. The idea behind reporting this-cases is to increase awareness of the above entity and for concurrent surgical treatment.

Keywords: Accessory and cavitated uterine mass, CPP, Dysmenorrhea

INTRODUCTION

Accessory and cavitated uterine mass (ACUM) represent a new variety of Mullerian anomaly. In 2010 Acién et al suggested the term ACUM as a new terminology and defined it by the presence of a non-communicating accessory uterine mass located in the myometrium or within the broad ligament, close to the round ligament insertion, with an otherwise normal genital and urinary tract.[1,2]

Since its first description by Cullen in 1908, different terminologies have been used to describe the same entity: juvenile or isolated cystic adenomyoma, uterus-like mass or accessory uterine cavity and adenomyotic cyst or cystic adenomyosis.[2-5]

Potter et al reported a case of a rare Mullerian anomaly in a young woman with non-communicating accessory uterine cavity adjacent to normal uterus.[9]

The criteria to diagnose ACUM (Acien et al):[1] An isolated accessory cavitated mass usually located under the round ligament. A normal uterus (endometrial lumen), fallopian tube and ovaries. A surgical case with an excised mass and pathological examination sharing an accessory cavity lined by endometrial epithelium with glands and stroma. Chocolate brown-coloured fluid as content. No adenomyosis (if the uterus has been removed). Although there could be small foci of adenomyosis in the myometrium adjacent to the accessory cavity.

According to Acién and his group, this anomaly required a separate classification and definition from the ESHRE 2013 consensus on congenital malformations of the female genital tract as that consensus does not include this anomaly. At the time of writing, it is considered as part of the unclassified uterine malformations (U6 class).[6]

In their opinion, the origin of this uterine anomaly could be a gubernaculum dysfunction during the embryogenesis expressed through a duplication and persistence of ductal Müllerian tissue at the attachment level of round ligament.[7]

Epidemiology

Prevalence of Mullerian anomalies ranges from 0.001-10% in general population and from 8-10% in women with adverse reproductive history.[10] Approximately 7% of young women have anatomical abnormality in their reproductive tract and the most frequent symptom of this is intolerable pain during period.[8]

CASE REPORT

The current preferred terminology is accessory cavitated uterine mass/or malformation (ACUM). We report two cases here within one year: I-36 years multiparous lady with complaints of right sided lower abdominal pain for 2 month not responding to medical management. II -25 years nulligravida who complained of severe dysmenorrhea after the onset of menarche, not responding to NSAIDs or hormonal medication.

Case 1

A 36 year old parous lady/previous 3 LSCS/came with complaints of right sided lower abdominal pain for past 2 months from march 2023 not relieved with NSAIDs and antispasmodics. H/O irregular menstrual cycles since 1.5 years, getting her cycles once in 3-4 months with normal flow for two days associated with dysmenorrhoea.

Abdominal examination

Soft, obese, LSCS scar healthy. On pelvic examination uterus 6-8 weeks size, mobile, right adnexa minimal tenderness present, POD free.

Transvaginal scan of pelvis

Uterus normal size, right ovary normal, right adnexal mass 4×4 cm seen, left ovary normal. Impression of rt adnexal mass? broad ligament fibroid? complex ovarian cyst.

Laparoscopy findings

Uterus of normal size. Bilateral fallopian tubes and ovaries normal. Intra operatively right adnexal solid mass 4×4 cm seen adjacent to the right uterine wall, lying separate from uterus in the broad ligament and below the insertion of round ligament. Mass was excised and removed into to along with rt fallopian tube in view of technical difficulty.

HPE finding

A nodule composed of endometrial glands in proliferative phase with stroma, foci of adenomyosis and hypertrophied myometrium.

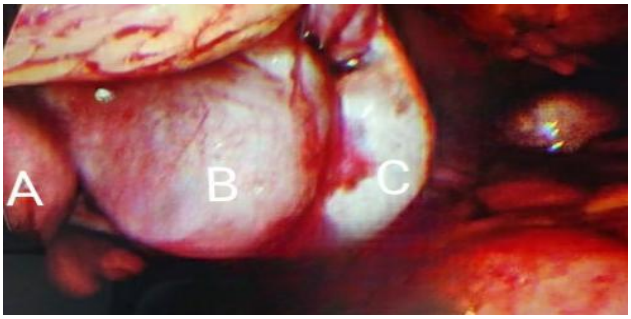

Figure 1 (A-C): Intraoperative findings showing, uterus, right adnexal mass and right ovary.

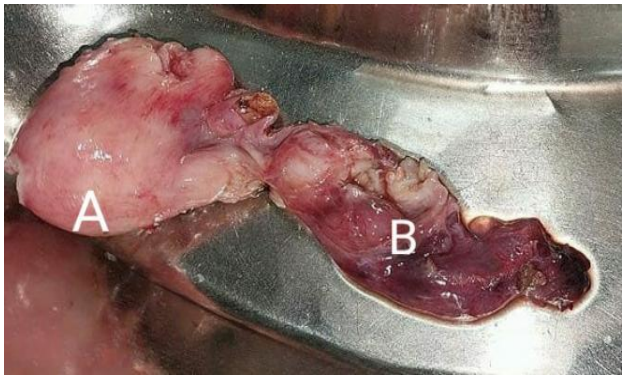

Figure 2: Postoperative specimen showing, right adnexal mass and right fallopian tube.

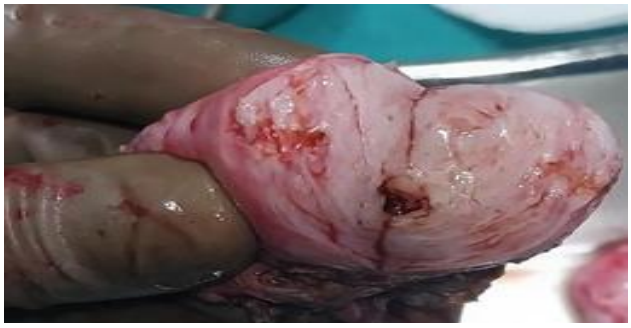

Figure 3: Right adnexal mass on cut section.

Figure 4: Solid right adnexal mass 4×4 cm with hypertrophied myometrium.

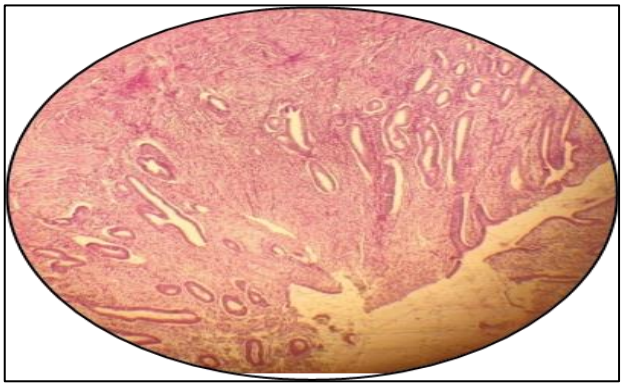

Figure 5: HPE of endometrial gland with lumen.

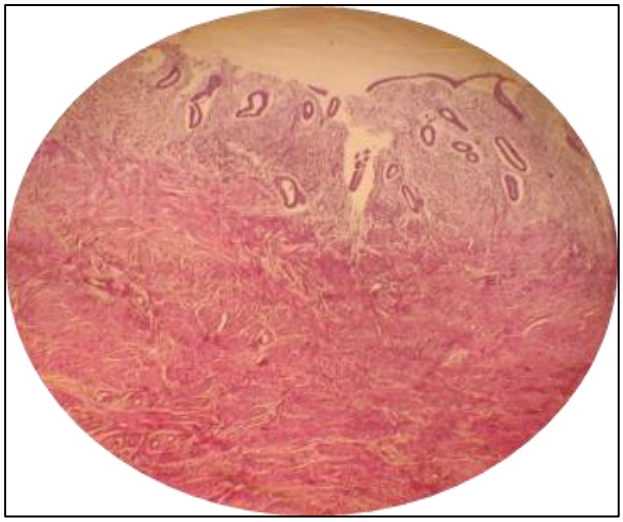

Figure 6: HPE showing foci of adenomyosis and hypertrophied myometrium.

Case 2

A 26-year-old nulligravida came with complaints of cyclical pain in the lower abdomen during and after periods since menarche for 13 years partly relieved with NSAIDs and OC pills. She was frequently hospitalized for the same and had to take iv analgesics. Her cycles were regular with normal flow. No bowel and bladder disturbances. H/O laparoscopic appendicectomy done in 2018 for pain in right iliac fossa. On examination abdomen was soft, not tender, lap appendicectomy scar present and healthy. She was evaluated with transabdominal scan which showed uterus of normal size, with hypoechoic area 2.1×1.7cm seen in the right lateral wall of the uterus. Central cystic area with echogenic lining measuring 1.2×0.9×1.4 cm (Vol-0.8 cc) not communicating with the uterine cavity. Endometrial thickness- 7mm. Both ovaries normal. Differential diagnosis of 1. Rudimentary horn with collection 2. Accessory cavitated uterine mass.

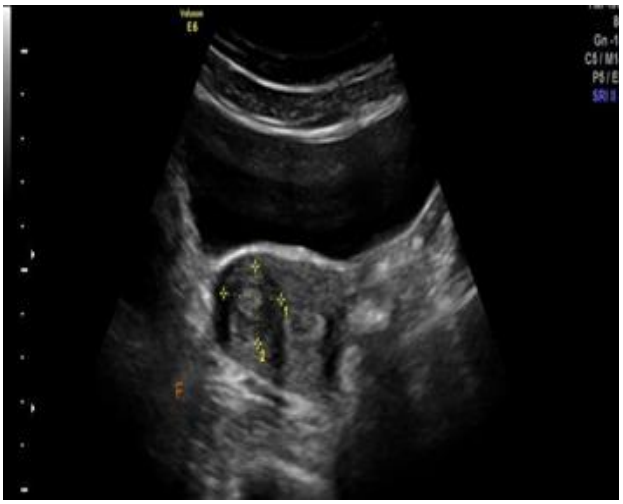

Figure 7: 2D Ultrasound showing uterus with endometrial cavity and an isolated cavitated mass.

Figure 8: 3D ultrasound showing isolated cavitated mass with collection in the Right lateral wall of uterus away from the morphologically normal uterine cavity.

She was planned for laparoscopic ACUM excision. Laparoscopic exploration revealed a mass of size 4×4 cm on the anterior and right lateral wall, lateral to the origin of round ligament and away from the tubal insertion. Mass was excised by subcapsular plane dissection after vasopressin injection. The mass which was present in the superficial myometrium was easily excised without entering the uterine cavity and without damaging bilateral tubes and round ligaments. Both ovaries normal. Uterus closed in layers with vicryl.

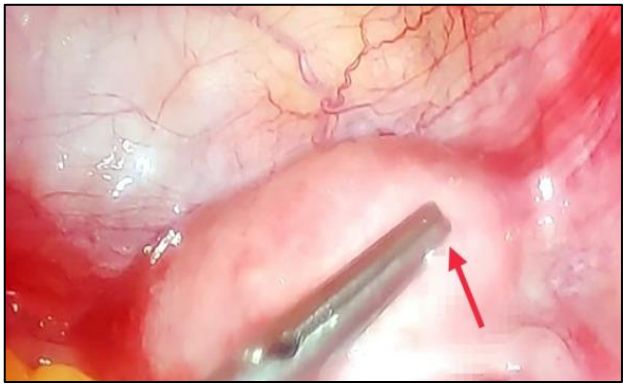

Figure 9: Arrow indicated right side accessory uterine mass below the round ligament.

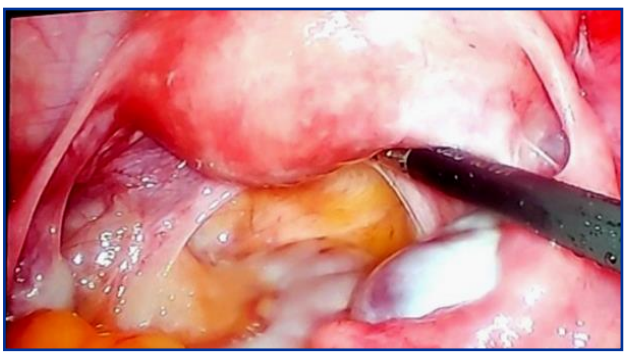

Figure 10: Normal bilateral tubes and ovaries, no adnexal collection in POD.

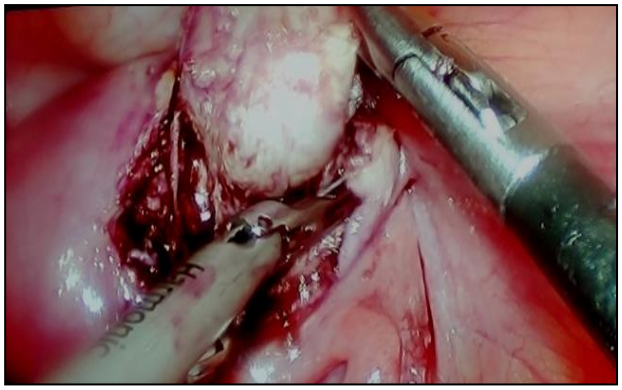

Figure 11: Right adnexal mass excision.

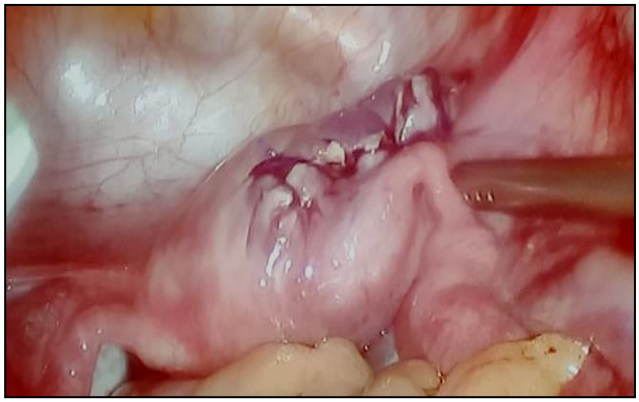

Figure 12: After excision of mass closed with polyglactin.

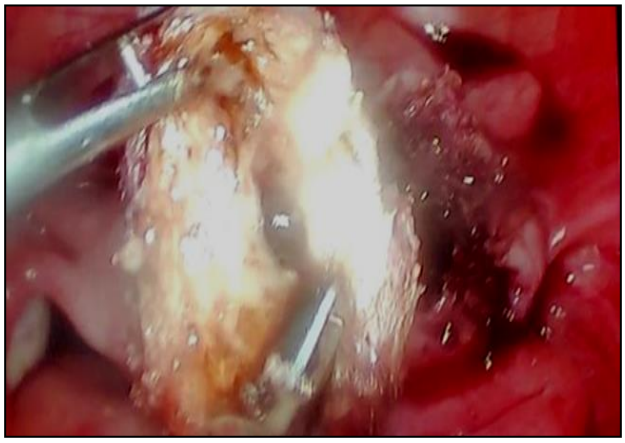

Figure 13: Accessory mass cut opened showing cavity and chocolate colour fluid inside.

On cut section cavity identified, inner surface was regular and chocolate coloured fluid drained.

Pathological findings

Specimen showed portion of smooth muscle tissue with areas of fibrosis. Blood vessels and nerve bundles are seen. No definite lining endometrium seen. No evidence of malignancy.

DISCUSSION

Case 1

With adnexal solid mass planned for diagnostic laparoscopy and proceed, uterine fibroid with degeneration was the probable diagnosis in this age group.

Most cases of ACUM have been diagnosed in women age less than 30 years and those who are nulliparous: however, some cases have been reported in women who are older than 30 year or are multiparous.[1]

In review of literature, focal cystic adenomyosis occurs in multiparous women of aged 35-40 years usually presented with heavy painful periods. On ultrasound ill-defined borders, located away from the junctional zone, with no significant mass effect on the endometrial cavity. The rest of the uterus may show features of focal or diffuse adenomyosis.[11] On histology surrounding myometrium lacks organization and absence of internal epithelial lining of cystic cavity is noted.[12]

ACUM presents with severe dysmenorrhea with ultrasound finding of isolated lesion in the lateral myometrium, inferior to the insertion of round ligament, separate and distinct from the endometrial myometrial junction.[13] On histology there was concentric organization of smooth muscle around cavity, endometrial glands and stroma seen to line the cavity.[14] This differentiates the ACUM from focal cystic adenomyosis which has definite endometrial gland with foci of adenomyosis and hypertrophied myometrium in this patient.

Degenerating fibroids are usually benign tumour of myometrial origin with cystic lesion in the middle of tumour and Fibroid can undergo various degeneration like hyaline, cystic, myxomatous and red degeneration. Histopathology findings of well circumscribed with solid rubbery firm texture.[15] It consisting of fascicles of elongated smooth muscles cells with eosinophilic cytoplasm and centrally located cigar shaped nucleus. It is rich in vasculature of various calibres and types including muscle rich arteries, arterioles and veins. Hence, the above findings differentiate ACUM from degenerating fibroid.

Case 2

Considering her symptoms of dysmenorrhea from menarche, we initially considered the differential diagnosis of a non-communicating rudimentary horn and treated symptomatically with antispasmodics. Inspite of analgesics she persistently presented to local hospital with pain where diagnostic laparoscopy+appendicetomy was done. Since the complaints recurred, she presented to our hospital again, since prior laparoscopic details not submitted patient was evaluated again and differential diagnosis of 1. ACUM 2. Rudimentary horn with collection 3. Bicornuate uterus.

Mullerian anomaly with uterine malformation are due to the failure of one of the mullerian duct to elongate towards the urogenital sinus while the contralateral Mullerian duct develops normally.[16] Usually associated with uterine malformation (unicornuate uterus, bicornuate uterus), 74-90% of unicornuate horn associated with rudimentary uterine horn.[17,18] An isolated cavity with horn and fallopian tube attaching on it without communicating with true cavity.

Here isolated cavitated cystic mass below the round ligament with normal shaped uterus and morphologically normal uterine cavity is one of the definite features of ACUM.[19] This is to differentiate ACUMs from obstructive and non-obstructive congenital uterine anomalies such as unicornuate uterus with a rudimentary horn or Robert's uterus which is defined as septate uterus with a non-communicating hemicavity. This patient has no uterine anomaly hence rudimentary horn with collection and bicornuate uterus were excluded from the differential diagnosis.

Diagnostic criteria for the ACUM were fulfilled except the histopathological findings of smooth muscle tissue with area of fibrosis and no definite lining of endometrium seen in our patient as specimen was morcellated for laparoscopic removal.

Severe dysmenorrhea is the commonest presenting symptoms for women with ACUM. The pain can be central or ipsilateral to the side of the ACUM and may be accompanied by CPP. The pain is thought to be caused by accumulation of menstrual fluid from the functioning endometrium lining the ACUM cavity. This would lead to increase pressure within the ACUM and subsequent stretching of the cavity. Pain often persists or even increases after the onset of menstruation. Other symptoms are dyspareunia and hypogastric pain. Some authors have described a clinically palpable, tender mass on bimanual vaginal examination, which could be mistaken for a fibroid or ovarian cyst.[19]

Follow up

Both patients came for follow up visits, no complaints of dysmenorrhea with regular cycles.

CONCLUSION

ACUM is now a well-defined uterine malformation with precise characteristics that every clinician should be aware of it. It should be a differential diagnosis in a patient of severe dysmenorrhea and CPP. ACUM is underdiagnosed because it is a poorly known entity, hardly ever researched in the context of acute and early dysmenorrhea not responding to medical treatment. The definite treatment is resection of the mass with preservation of fertility in young nulliparous women.

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Ethical approval: Not required

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