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Twin pregnancy in an accessory cavitated non-communicating uterus



Harith M. Alkhateeb*, Enas M. Yaseen

College of Medicine, Tikrit University, Iraq

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ABSTRACT

BACKGROUND: A uterine malformation is a type of female genital malformation resulting from abnormal development of the Mullerian duct(s) during embryogenesis. The type and degree of uterine malformation depends on the level at which the fusion process of the two Mullerian ducts stops; thus, there is a wide variety of malformations. A newly described deformity called accessory cavitated uterine mass (ACUM) has been increasingly reported.

THE CASE: We report this deformity (in a 20-year-old married woman) which appears to be an additional incompletely developed, cavitated and presumably non-communicating uterus in addition to a normally shaped and developed uterus. The former uterus became impregnated with twins that died in a missed abortion at 13 weeks of gestation. Before discovering the presence of the deformity, three attempts were made to evacuate the dead fetuses by cervical dilatation and curettage of the normal empty uterus. These attempts resulted in perforation of its fundus, a laparotomy was performed to repair the uterus. During the laparotomy, the pregnant accessory uterus was discovered and was excised with the dead twins.

DISCUSSION: The lack of good medical history was a cause of the mismanagement of this patient. Most probably, the origin of ACUM is a growth from the right Mullerian duct. The ovum has entered the ACUM through the rudimentary tube and has been fertilized by a sperm travelled either through the normal vaginal and uterine cavities or through the lymphatics.

CONCLUSIONS: (1) A detailed case history is important. (2) An ACUM can be impregnated.

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1. Introduction

Uterine malformations can cause amenorrhea, infertility, recurrent pregnancy loss and pain and can also be normally functioning, depending on the nature of the defect [1–3,5]. The prevalence of uterine malformation is estimated to be 6.7% in the general population, slightly higher (7.3%) in the infertile population, and significantly higher in a population of women with a history of recurrent miscarriages (16%) [4,6].

Until the sixth week of life, the male and female genital systems are identical.

There are two pairs of symmetrical genital ducts, the mesonephric (Wolffian) ducts and the paramesonephric (Mullerian) ducts [1,2,7].

In the female embryo, the mullerian ducts grow caudally and become enclosed in the peritoneal folds, which later give rise to the broad ligaments of the uterus to which the ovaries, fallopian tubes and uterus are attached. The Mullerian ducts approach each other and begin to fuse. At 9 weeks gestation, the septum from the fused ducts begins to resorb. This becomes the uterus and the upper portion of the vagina [2,7,8].

Mullerian tract anomalies result from incomplete bilateral duct elongation, fusion, canalization, or septal resorption of the Mullerian ducts. These anomalies may occur in any step during this developmental process [1,3,9].

An accessory cavitated uterine mass (ACUM) is associated with an otherwise normal uterus. ACUM can be difficult to differentiate from true cavitated adenomyomas and cavitated rudimentary uterine horns. An accessory uterine mass can be caused by duplication and persistence of the ductal Müllerian tissue in a critical area at the attachment level of the round ligament, possibly related to gubernaculum dysfunction. For the differential diagnosis of ACUM and cavitated non-communicating rudimentary uterine horns, hysterosalpingography showing a normal eutopic uterine cavity is helpful [10,11].

Abbreviations: U/S, ultrasound; HSG, hysterosalpingography; ACUM, accessory cavitated uterine mass; ov, ovary.

* Corresponding author.

E-mail addresses: khateeb_hm@yahoo.com (H.M. Alkhateeb), enasyaseen51@yahoo.com (E.M. Yaseen).

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An old classification had been put by Jone H.W Jr. in 1953 for the congenital anomalies of the female urogenital system [12]:

1. Uterus: (a) Single uterus. (b) Septate uterus. (c) Bicornuate uterus. (d) Double uterus.
2. Cervix: (a) Single cervix. (b) Septate cervix. (c) Double cervix.
3. Vagina: (a) Single vagina. (b) Septate vagina. (c) Double vagina.

A recent ESHRE/ESGE classification of female genital tract anomalies [13]

(1) Uterine anomalies:

- U0. Normal uterus.
- U1. Dysmorphic uterus: (a) T-shaped. (b) Infantalis. (c) Others.
- U2. Septate uterus: (a) partial. (b) Complete.
- U3. Bicornuate uterus: (a) partial. (b) Complete. (c) Bicornuate uterus.
- U4. Hemi-uterus: (a) with rudimentary cavity. (b) Without rudimentary cavity.
- U5. Aplastic: (a) with rudimentary cavity (bi- or unilateral horn). (b) Without rudimentary cavity (bi- or unilateral uterine remnants/aplasia).
- U6. Unclassified malformations.

• Cervical anomalies:

- C0. Normal cervix.
- C1. Septate cervix.
- C2. Double normal cervix.
- C3. Unilateral cervical aplasia.
- C4. Cervical aplasia.

• Vaginal anomalies:

- V0. Normal vagina.
- V1. Longitudinal non-obstructing vaginal septum.
- V2. Longitudinal obstructing vaginal septum.
- V3. Transverse vaginal septum and/or imperforate hymen.
- V4. Vaginal aplasia.

2. The case

A 20-year-old female married for 2 years had undergone a full-term pregnancy with normal vaginal delivery, one year prior to this case report. The patient presented with 3 missed periods with lower abdominal pain, and a pelvic ultrasound examination revealed an intrauterine pregnancy of twins that had died at 13 weeks of gestation (miscarriage) (Fig. 1). Induction of abortion by oxytocin drip had been attempted twice but failed. The gynecologist proceeded to perform cervical dilatation and curettage, but the uterus was found to be empty. A post curettage ultrasound showed that the miscarried twins were still present. The patient was seen by another gynecologist who repeated the curettage and also did not find the dead twins, and an ultrasound after the procedure still showed the dead twin fetuses. The patient was then seen at our clinic, and we submitted the patient to another curettage. We observed that the omental tissue had pulled out through the vagina. Perforation of the uterus was expected; thus, we opened the abdomen and found a slightly enlarged normally shaped and positioned uterus with 2 fallopian tubes and 2 ovaries. There was a uterine perforation at the fundus; thus, the omentum was pulled in through the perforation, drawn out of the uterus, transfixed, ligated by suture and trimmed. The perforation of the fundus was repaired with 3 stitches using one zero vicryl sutures, no other visceral injury was found.



Fig. 1. An ultrasonogram of the gravid accessory uterus showing dead twins of 13 week + 2 days gestation marked as FETUS 1 and FETUS 2.

On exploration, we found a large pear-shaped mass emerging from the right broad ligament with a thick pedicle that ended near the uterine cervix. Because we did not obtain a detailed gynecological history of the patient, we assumed the mass was a broad ligament cyst. The mass was easily excised by clamping, transfixing, ligating and cutting the pedicle. It was 2 cm in width and fleshy in nature.

3. Surgical pathology

The mass is pear-shaped and 10 × 13 cm in size. The wide upper portion of the mass has 2 small appendages at either corner that are each 1 cm long (Fig. 2). The narrow lower end of the mass was firm in consistency (arrow in Fig. 2).

We opened the mass and observed amniotic fluid and two dead fetuses, each 6 cm in length (Fig. 2). The mass wall was thin in the upper segment and thick in the lower segment. The portion of the mass representing the cervix was solid and did not have an internal opening or cervical canal, as confirmed using a fistula probe.

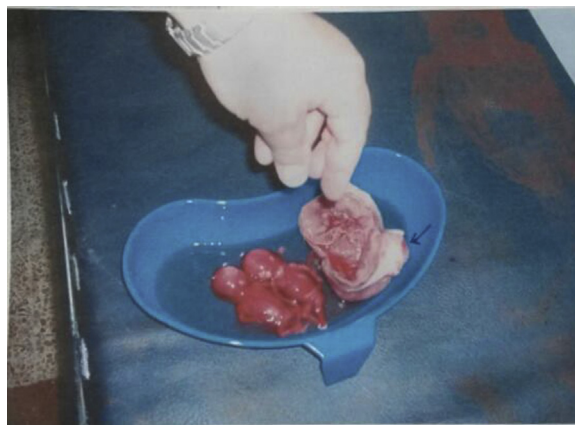


Fig. 2. The excised accessory uterus opened through its posterior wall to the fundus showing the uterine cavity containing an amniotic sac and the two dead fetuses outside with evident umbilical cords. An arrow pointing to the blind cervical canal.

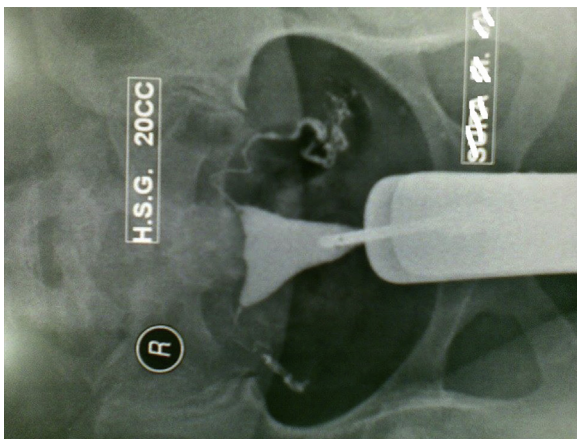


Fig. 3. A post-operative hysterosalpingogram showing a normal uterine cavity with two patent fallopian tubes. The name of the patient on the X-ray film is obscured by white lines for privacy purposes.

Tissue samples taken from the upper and lower portions of the specimen for histopathological analysis both exhibited the smooth muscle fiber arrangement of uterine tissue.

The patient had a smooth post-operative period. One year later, a hysterosalpingogram showed a normal uterine cavity and two patent fallopian tubes (Fig. 3). A few months later, she became pregnant, and she had a full term pregnancy with normal vaginal delivery. She had four subsequent pregnancies over 9 years that all reached full term and all resulted in normal vaginal deliveries.

4. Discussion

There was a lack of a full medical history and poor communication between the gynecologist and the ultrasonographer. Thus, despite the proficiency of the ultrasonographer, during two examinations, he did not thoroughly examine all pelvic organs once he detected a twin miscarriage in what appeared to be a uterine cavity. If he had examined all pelvic organs, he should have detected the empty normal uterus just beside the gravid abnormal uterus as it is clear in the post-operative hysterosalpingography (Fig. 3). Hysteroscopy could have been useful in this case, but unfortunately, the only available hysteroscope in the hospital was broken-down.

Most of the congenital anomalies of the uterus are due to a fault in one or more step in the fusion process of the Mullerian ducts. In this case, however, the accessory uterus most probably developed as a growth from the right Mullerian duct generating an incompletely developed separate accessory uterus (non-communicating, cavitated accessory uterus) ACUM [10,11].

Most probably, the ovum entered the accessory uterus through the small appendages on the sides of the dome, which represent rudimentary fallopian tubes. The ovum likely became fertilized by sperms passing through the normal cervical canal of the normal uterus, uterine cavity, fallopian tube, and the peritoneal cavity, or the sperms may have travelled through the lymphatic vessels of the accessory uterus to fertilize the ovum inside it [14].

5. Conclusions

1. A deficient case history, lack of communication between the clinicians and the ultrasonographer and repeated shifting of the patient from one doctor to another, all led to the misdiagnosis and mismanagement of this patient.
2. Conception can occur in ACUM, however, it is very rare and may be difficult to diagnose.

Competing interests

The authors declare that they have no competing interests.

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Ethical Approval

It is not a research study.

Author's contributions

Harith M. Alkhateeb: surgery for the patient, writing of original text, communication with the editorial team.

Enas M. Yaseen: the last gynecologist dealing with the patient, periodical clinical examinations and follow up of the patient.

Consents

Written informed consents were obtained from the patient and her husband. A copy of the written consents are available for review by the Editor-in-Chief of this journal.

Guarantor

Harith Mustafa Alkhateeb

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References

- [1] L. Change, D. Muran, *Pediatric and Adolescent Gynecology: Current Obstetric and Gynecological Diagnosis and Treatment*, in: A.H. Decherney, L. Nathan, N. Laufer, A.S. Roman (Eds.), 11th ed., Lange, 2012, pp. 38–66.
- [2] A.L. Nelson, J.C. Gambone, *Congenital anomalies and benign conditions of uterus*, in: I.M. Symond, S. Arulkumaran (Eds.), *Essential Obstetrics and Gynecology*, 5th ed., Churchill Livingstone, 2010, pp. 240–247.
- [3] *Gynecology by Ten Teachers*, in: A. Monga, S. Dobbs (Eds.), 19th ed., Hodder Arnold, UK, 2011, pp. 23–24.
- [4] *Obstetric by Ten Teachers*, in: P.N. Baker, L.C. Kenny (Eds.), 19th ed., Hodder and Stoughton, 2011, p. 88.
- [5] D.K. Edmonds, *Normal and abnormal development of the genital tract*, in: *Deuhartst's Text book of obstetrics and Gynecology*, 8th ed., Wiley-Blackwell, 2011, pp. 421–432.
- [6] P. Acién, F. Sánchez del Campo, M.J. Mayol, M. Acién, *The female gubernaculum: role in the embryology and development of the genital tract and in the possible genesis of malformations*, *Eur. J. Obstet. Gynecol. Reprod. Biol.* 159 (2011) 426–432.
- [7] C. Cozzutto, *Uterus-like mass replacing ovary: report of a new entity*, *Arch. Pathol. Lab. Med.* 105 (1981) 508–511.
- [8] H. Takeuchi, M. Kitade, I. Kikuchi, J. Kumakiri, K. Kuroda, M. Jinushi, *Diagnosis, laparoscopic management, and histopathologic findings of juvenile cysticadenomyoma: a review of nine cases*, *Fertil. Steril.* 94 (2010) 862–868.
- [9] M.A. Bedaiwy, D.N. Henry, S. Elguero, S. Pickett, M. Greenfield, *Accessory and cavitated uterine mass with functional endometrium in an adolescent: diagnosis and laparoscopic excision technique*, *J. Pediatr. Adolesc. Gynecol.* 26 (2013) e89–91.
- [10] P. Acién, M. Acién, F. Fernández, M. José Mayol, I. Aranda, *The cavitated accessory uterine mass: a Müllerian anomaly in women with an otherwise normal uterus*, *Obstet. Gynecol.* 116 (2010) 1101–1109.
- [11] P. Acién, A. Bataller, F. Fernández, M.I. Acién, J.M. Rodríguez, M.J. Mayol, *New cases of accessory and cavitated uterine masses (ACUM): a significant cause of severe dysmenorrhea and recurrent pelvic pain in young women*, *Hum. Reprod.* 27 (2012) 683–694.

- [12] P. Acien, M.I. Acien, The history of female genital tract malformation classifications and proposal of an updated system, *Hum. Reprod. Update* vol. 17 (No. 5) (2011) 693–705.
- [13] G.F. Grimbizis, S. Gordts, A. Di Spezio Sardo, S. Brucker, C. De Angelis, M. Gergolet, T.-C. Li, V. Tanos, H. Brolmann, L. Gianaroli, R. Campo, The ESHRE/ESGE consensus on the classification of female genital tract congenital anomalies, *Hum. Reprod.* vol. 28 (No. 8) (2013) 2032–2044, Downloaded from: <http://humrep.oxfordjournals.org/> by guest on June 22, 2014.
- [14] J. Mazin Khasbag, Spermatozoal migration and lymphatics: a hypothesis, *Med. Hypothesis* vol. 2 (November and December (6)) (1976) 275–276.

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