

Accessory uterine appendage may be a new müllerian malformation

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Objective: To describe a case of an accessory uterine structure.

Design: Case report.

Setting: University affiliated, community-based hospital.

Patient(s): A 35-year-old woman who presented for laparoscopic tubal sterilization.

Intervention(s): Laparoscopy, tubal sterilization, dye test, and excision of accessory uterine structure.

Main Outcome Measure(s): Laparoscopic findings and resection of the anomaly.

Result(s): Laparoscopy showed a hollow uterine appendage that communicated with the uterine cavity. This was completely excised.

Conclusion(s): We report a rare case of a uterine anomaly that is inconsistent with the traditional classification system, and we offer a possible mechanism for its formation. (*Fertil Steril*® 2005;84:1017.e7–9. ©2005 by American Society for Reproductive Medicine.)

Key Words: Müllerian, uterine anomaly, uterine malformation

The reported prevalence of congenital uterine anomalies ranges from <1% to 27%, depending on the population studied and the method of diagnosis (1, 2). A retrospective review of 3,181 patients in whom the morphologic features of the uterus were ascertained by hysterosalpingogram and laparoscopy/laparotomy found that the overall frequency of uterine malformations was 4% (3). In this review, the prevalence was higher in patients with two or more consecutive pregnancy losses (6.3%) compared with fertile patients (3.8%) and patients with failure to achieve pregnancy for >2 years (2.4%). Another review of women with normal reproductive outcome in whom the morphologic features of the uterus were ascertained by laparoscopy/laparotomy and hysterosalpingogram found uterine malformations in 3.2% (4).

In 1988, the American Society for Reproductive Medicine published a classification for müllerian duct malformations, based on the embryologic development of the reproductive tract (5). However, several case reports have identified utero-vaginal malformations that do not fit into any of these diagnostic categories. Quagliarello et al. (6) reported a case in which an accessory cervix was present, with a normal uterus and endometrial cavity and normally positioned fallopian tubes. There have been reports of septate uteri with cervical duplication, and these do not fit into the current classification (7, 8). Potter and Schenken (9) reported a case of a noncommunicating accessory uterine cavity, with both fallopian tubes communicating with the main uterine cavity.

Some of these malformations call into question the embryologic theory of caudal to cranial fusion of the müllerian ducts. We present an unusual uterine malformation, which to our knowledge has never been reported before.

CASE REPORT

A 35-year-old woman presented for laparoscopic tubal sterilization (LTS). She had had two previous vaginal deliveries at term, and two previous first trimester spontaneous miscarriages. Her medical, surgical, and family history were unremarkable. A 6-week pelvic ultrasound performed during her first pregnancy in 1996 did not document any uterine abnormality.

At the time of the laparoscopic tubal sterilization, no abnormality of the vulva, vagina, or cervix was noted. The uterus was noted to be normal in shape and size, with a normal fallopian tube on either side. The round ligaments on both sides also appeared normal.

At the uterine fundus, slightly to the right of the midline, were two tubular structures, extending from the fundus. Each structure was 2-cm long, and both extended into a sac-like structure measuring 1 × 1 cm. From the opposite end of this sac-like structure, there was another 3-cm tubular structure, which ended blindly. This accessory uterine structure was freely mobile (Fig. 1).

Tubal sterilization was performed by cauterizing the normally sited fallopian tubes. Indigo carmine dye was then injected through the cervix. No spillage of dye was seen through the cauterized fallopian tubes. The accessory uterine structure (AUS) filled with dye, but there was no spillage. Each base of the tubular attachments of the AUS to the

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FIGURE 1

Uterus with an accessory uterine appendage (*arrowhead*) extending from the fundus.



Umobi. Accessory uterine appendage. Fertil Steril 2005.

uterine fundus was cauterized, and it was excised. The dimensions given above were obtained on measurements, and it was sent for pathologic examination. The histopathology report read, “orderly smooth muscle bundles with evidence of remote hemorrhage and superficial telangiectasia. No tumor noted.”

The patient was asked to get a postoperative intravenous pyelogram, but she declined.

DISCUSSION

The uterus is formed from fusion of the caudal portions of both müllerian ducts. The cranial portions remain unfused to form the fallopian tubes. There are seven classifications of uterine anomalies proposed by the American Society for Reproductive Medicine (2). Some investigators have used presumed causative mechanisms to group these malformations, such as disorders of lateral fusion (obstructive or nonobstructive) and vertical fusion (obstructive or nonobstructive) (10–12).

The uterine anomaly in this case does not appear to fit into any of these categories, and we offer a possible mechanism for its formation. In our patient, the normal uterus, with a normally sited fallopian tube on each side, suggested that the müllerian ducts from both sides had fused

normally. The AUS was hollow, with a cavity that communicated with the main uterine cavity as evidenced by its filling with dye. This suggested a possible diverticular-like mechanism, leading to an outgrowth from the müllerian ducts. The AUS was attached to the uterus via two tubular structures, which probably originated from either müllerian duct. These anomalous outgrowths became subsequently fused to one another in the same manner as the original müllerian ducts.

We had tried to do a thorough review of the histopathology of the AUS, but the original slides or specimen were no longer available. Because this was an incidental finding at the time of laparoscopic tubal sterilization, the patient never had a preoperative hysterosalpingogram. We therefore cannot comment on the configuration of the uterine cavity with certainty. The only information we have to suggest a normal uterine cavity is her previous first trimester transvaginal ultrasound. Ultrasound has a reported sensitivity of 57% to 100% for detecting müllerian tract abnormalities (13, 14). We also cannot comment on the presence or absence of any associated renal malformation, as an intravenous pyelogram was not performed as stated above.

In conclusion, we report a rare case of a uterine anomaly that is inconsistent with the traditional classification system, and we offer a possible mechanism for its formation.

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