Management of a large uterine cyst protruding through the cervix of a mare

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Abstract

A 17-year-old, multiparous, American Quarter Horse mare was presented for breeding 7 days after foaling. Transrectal ultrasonography revealed multiple endometrial lymphatic cysts; 2 at the base of the left uterine horn and 1 in the caudal uterine body that extended into the cervical lumen (the latter was surgically removed). The mare was artificially inseminated on the subsequent estrous cycle, confirmed in foal 30 days after ovulation and delivered a viable foal without complications 350 days after ovulation.

Keywords: Lymphatic cyst, cervix, endometrial cyst, endometrium

Background

A multiparous, American Quarter Horse mare was presented for artificial insemination at Equine Reproduction Services at Kansas State University in 2016. Her last foal was born in 2015 and was diagnosed not-pregnant following artificial insemination that season. During the 2016 breeding season, a breeding soundness examination was performed. During an ultrasonographic examination, 2 endometrial lymphatic cysts were detected at the base of the left uterine horn; 1 was 15.7 x 8.7 mm and the other 9.6 x 9.0 mm. Endometrial cytology and culture were performed. A non-inflammatory cytology and a mixed population of bacteria (*Streptococcus equinus, Streptococcus dysgalactiae*, and *Actinobacillus* sp.) considered non-pathogens were recovered on enrichment only. No other abnormalities were detected on physical or vaginal examination or on transrectal palpation ultrasonography. On the following estrous cycle, the mare was given deslorelin acetate (Sucromate™, Thorn BioScience, Louisville, KY), and then artificially inseminated once with fresh, cooled semen. Using transrectal ultrasonography, the mare was confirmed in foal 16 days after ovulation. She was maintained at pasture with other brood mares and supplemented with grass hay and commercial feed. The mare had an unattended foaling on May 24, 2017 at 331 days gestation. The owner reported that the foal was found dead enclosed within the amnion. Neither the fetus nor fetal membranes were examined or submitted for evaluation.

Case presentation

The mare presented with a good body condition score (5/9) 7 days after foaling for breeding during foal heat. Her perineal conformation was within normal limits and no vaginal discharge was present. Transrectal palpation revealed a postpartum uterus that was involuting and of expected size for 7 days postpartum. Transrectal ultrasonography revealed the presence of the previously detected endometrial lymphatic cysts at the base of the left uterine horn with no change in size. However, a large fluid-filled mass (Figure 1) with mixed echodensity, 35.7 x 44.7 mm, was present at the caudal uterine body and projected into the cervical canal. A tail wrap was placed and the perineum cleansed with a gloved hand and cotton using warm water and a mild detergent soap. Digital examination of the cervix revealed a fluid-filled lobulated structure originating within the caudal uterine body and extending into the cervical canal. The mass was attached to the dorsal wall of the uterine endometrium in a sessile manner with a broad fibrous base approximately 50 mm long and could not be reduced back into the uterine body. The mare was sedated with 0.01 mg/kg detomidine hydrochloride IV (Dormosedan®, Zoetis Inc., Kalamazoo, MI). Hysteroscopy was done with a cold-sterilized video endoscope, confirming the presence of a mass in the cervical canal and caudal uterine body.

Differential diagnosis

The differential diagnosis considered for a fluid-filled mass in this mare included a large endometrial lymphatic cyst, hematoma, abscess and neoplasia. Endometrial cysts are commonly described as having anechoic fluid or appearing hypoechoic on transrectal ultrasonography.[1] In this case, the mixed

echodensity of the fluid and the hyperechoic structure within the fluid were not typical for an endometrial lymphatic cyst. Furthermore, the location of the mass at the caudal uterine body and within lumen of the cervix was atypical, as endometrial cysts are more commonly located at the base of uterine horns.[2] Based on the mixed echodensity, location, and recent foaling, it was hypothesized that the mass could be a hematoma or an abscess secondary to trauma during foaling. Although uterine neoplasia (leiomyoma, leiomyosarcoma, fibroleiomyomas, and adenocarcinoma) was considered as an initial differential, it was considered unlikely, as the mass did not have the typical consolidated, hyperechoic appearance on transrectal ultrasonography and hysteroscopy that would be expected with any of the uterine neoplastic conditions.[1]

Treatment

The mare was sedated with 0.01 mg/kg detomidine hydrochloride IV (Dormosedan®, Zoetis Inc.,). A tail wrap was placed and the perineum cleansed with a gloved hand and cotton, using warm water and a mild detergent soap. The uterine mass was removed using a snare technique.[3,4] Approximately, 150 cm of obstetric wire (OB Saw Wire, Jorgensen Laboratories, Loveland, CO) was passed through two sterilized artificial insemination pipettes to create a loop between them. A sterile obstetrical sleeve with sterile lubricant was placed on one arm and passed along with the loop through the vagina. The loop was placed around the mass, and tightened by pushing the pipettes inward against the base of the mass tightening the wire. The wire was then moved back and forth through the pipettes cutting through the base of the mass and the mass was subsequently removed *per vaginum*.[3,4] Once removed, a lobulated, fluid-filled mass measuring approximately 70 mm long and 50 mm in diameter was submitted for histopathology and cytology. Uterine lavage with 2 liters of sterile lactated Ringers solution was performed immediately following the mass removal and again the following day with 1 liter sterile lactate Ringers solution. Each uterine lavage was followed by 20 USP units of oxytocin IM (Oxytocin, Bimeda, Oakbrook Terrace, IL). The mare was given 1.1 mg/kg flunixin meglumine IV (Banamine®, Merck Animal Health, Madison, NJ) following mass removal and prescribed 2 grams phenylbutazone paste (Phenylbute® Paste, Phoenix Pharmaceutical, St, Joseph, MO) to be given orally once daily for 2 days, starting the following day.

Diagnostic testing

The mass was submitted to Kansas State University Veterinary Diagnostic Laboratory (1800 Denison Ave, Manhattan, KS) for fluid analysis and histopathology. The fluid had low nuclear cellularity, with a low density of erythrocytes, occasional extracellular pink proteinaceous globules and pale basophilic backgrounds with multiple protein crescents. Nucleated cell populations consisted of few nondegenerative neutrophils and foamy macrophages, some of which contained phagocytized protein material. No infectious agents were observed and was reported as a mixed cell infiltrate with protein debris supportive of cystic material. Histopathology revealed a roughly round cystic structure composed of a large central cavity lined by attenuated cells that resembled lymphatic endothelium (Figure 2). The cystic cavity contained eosinophilic fibrillary debris with scattered moderate numbers of macrophages, lymphocytes, and plasma cells that compressed the surrounding endometrial tissue (which was moderately to markedly edematous). There were multifocal areas of mild to moderate hemorrhage within the surrounding endometrial mucosa. Throughout the mucosa and submucosa of the endometrium there was moderate to marked dilation of lymphatics and scattered inflammatory infiltrates of lymphocytes and plasma cells. These histopathologic findings were most consistent with an endometrial lymphatic cyst.

Outcome

The mare ovulated the day after removal of the endometrial lymphatic cyst. Five days after ovulation, cervical tone and function were assessed via vaginal examination and digital palpation. No abnormalities were detected, but potential for cervical adhesion formation was discussed with the owner, as well as incompetent cervical function throughout gestation, predisposing to placentitis. As the mare owner elected to rebreed the mare, 10 mg dinoprost tromethamine (Lutalyse®, Zoetis Inc.,) was given IM.

Deslorelin acetate (Sucromate™, Thorn BioScience, Louisville, KY) was given to the mare while in estrus, with a 35 mm follicle present. The mare was artificially inseminated with 1 dose of fresh, cooled semen 12 days after cyst removal (21 days after foaling) with ovulation confirmed the following day. The mare was monitored for the presence of mating induced endometritis with transrectal ultrasound prior to discharge. She was confirmed in foal 14 and 30 days after ovulation with transrectal ultrasonography. A Caslick procedure was performed following confirmation of pregnancy on day 14. Recommendations were made to evaluate the mare's pregnancy with transrectal palpation and transrectal ultrasonography at 60 - 70 days of gestation, and at monthly intervals starting at 7 months gestation to monitor for signs of placentitis. Further recommendations were made to vaccinate the mare for equine herpes virus 1 at 5, 7, and 9 months in pregnancy and to give other pre-foaling vaccinations. In addition, she was to have the Caslick opened 4 - 6 weeks prior to her expected due date. At 350 days after ovulation, the mare delivered a viable foal without assistance.

Discussion

Endometrial lymphatic cysts have been reported in a variety of species, including horses, cattle, ewes, pigs, cats, dogs, elephants and humans.[5] Uterine cysts are best described as anechoic, fluid-filled endometrial structures filled with yellow, watery fluid that generally increase in size over months to years. They may be seen as an individual, compartmentalized sacs, or as multilobulated structures divided by septa. They are often pedunculated, but may also be attached by a sessile base.[5] If large enough or numerous they may be detected via transrectal palpation, but are best characterized with transrectal ultrasonography. The incidence of uterine cysts has been reported to be 22 - 55%, with an increasing incidence with mare age and parity.[5-7] Uterine cysts are more likely in mares 10 years of age or older, but have been detected in a mare as young as 3 years.[6] In 1 study, mares > 11 years of age were 4.2 times more likely to have uterine cysts than mares < 11. In an earlier study with 259 mares, 22.4% had uterine cysts, although 73.1% of the uterine cysts were in mares > 14 years.[7] In another study with 76 mares, 44.7% had uterine cysts, with 2 mares < 7 years of age, 12 were from 7 - 14 years, and 20 were > 14 years.[5] Number of cysts was not different between left versus right horns, but frequency of cysts was higher in area of vesicle implantation (left horn middle, right horn middle, uterine body middle, cornual-body junction, left horn posterior, right horn posterior, and anterior uterine body) compared to the extremities (left horn anterior, right horn anterior, and posterior uterine body).[5,7] Two types of uterine cysts have been described, namely glandular and lymphatic.[2,3,5]

Glandular cysts, located in the lamina propria, are generally small, ranging from only a few millimeters up to 1 cm in diameter.[5,6] These glandular cysts develop from distention of endometrial glands due to chronic endometrial changes and more specifically periglandular fibrosis.[1,4-6] Glandular distention then occurs from the strangulating effect of the periglandular fibrosis on endometrial glands and decreased uterine myometrial tone resulting in decreased flow of secretions.[6] Glandular cysts have been reported as a normal finding during pregnancy.[5,6] The significance of glandular cysts in cases of infertility or subfertility is yet to be defined.

Lymphatic cysts, the most common form of uterine cysts, likely have more clinical relevance for fertility when compared to glandular cysts. Lymphatic cysts, or pooling of lymphatic fluid, initially develop in the stroma of the endometrium as microscopic structures from obstructed lymphatic channels or potentially from the gravitational effects of an enlarged gravid uterus, or postpartum uterus.[5-7] As they further develop and enlarge, due to continued pooling of lymphatic fluid, they distend and enter into the uterine lumen. Diameters of lymphatic cysts have been reported to range between 2 - 48 mm, and appear to increase in size with age of the mare.[5-7] Clinically, lymphatic cysts may or may not have an impact on pregnancy rate and foaling rate. The presence of lymphatic cysts may make initial pregnancy detection challenging as they may be confused with an early embryonic vesicle, or they may make the loss of pregnancy difficult to confirm. The clinical impact of endometrial lymphatic cysts is dependent on their size, number and location. It was reported that when the number of cysts was > 5, there was a negative correlation with foaling rate.[6] Endometrial cysts may negatively affect fertility by impeding mobility of the embryonic vesicle and thus blocking maternal recognition.[7] Furthermore, direct contact between the

embryonic vesicle and the cyst, instead of endometrium, could negatively affect nutrient absorption by reducing the area of total placental contact and nutrient exchange.[7] Pregnancy rate at day 40 in mares without uterine cysts was 88% compared to 71.4% in mares with uterine cysts,[7] although embryo loss between days 14 and 40 was not significantly different between the 2 groups.[7]

Using transrectal ultrasonography, endometrial cysts should be mapped out and their size and location documented. Depending on the number, size and location of endometrial cysts clinicians may elect to not initiate treatment or cyst removal, although removal of large or multiple cysts may increase fertility.[3] Multiple methods of cyst removal have been described, including a snare, laser hysteroscopy, loop cautery, and uterine biopsy or needle aspiration.

The location of the caudal uterine body and involvement of the cervical canal was not a typical finding of endometrial lymphatic cysts. In the present case, it is unknown when the endometrial cyst at the level of the caudal uterine body and cervical canal developed, although it either developed during pregnancy or rapidly postpartum. It was not present or detectable via transrectal ultrasound when the mare was managed for breeding in 2016. Due to delivery of a nonviable foal in 2017, it is possible that it developed during the pregnancy and jeopardized the pregnancy by affecting cervical function, perhaps predisposing the mare to an ascending placentitis. It is also possible that it rapidly developed and extended into the lumen after foaling. In order to restore cervical function and attempt to allow the mare to successfully carry a pregnancy to term, surgical removal was performed, carefully trying to not remove any portion of the cervical epithelium as the cyst was removed from the caudal uterine body. Due to its proximity to the cervix, main complications considered following removal of the cyst included cervical adhesions leading to a future dystocia, inability of the cervix to function properly predisposing the mare to subfertility, early embryonic death, or placentitis. Other complications considered less likely were intrauterine adhesions or cyst reoccurrence. If cervical adhesions were to occur, they were more likely to be linear and much less likely to be circumferential which should result in a lower occurrence of dystocia compared to circumferential adhesions.

Learning points

- Lymphatic cysts may make initial pregnancy detection challenging, as they may be confused with an embryonic vesicle, or a non-viable embryo.
- The clinical impact on fertility of endometrial lymphatic cysts is dependent on their size, number, and location.
- Presence of > 5 endometrial lymphatic cysts can reduce foaling rate.
- Removal of the cyst is possible by a snare and methods such as laser hysteroscopy, loop cautery, uterine biopsy, or needle aspiration.

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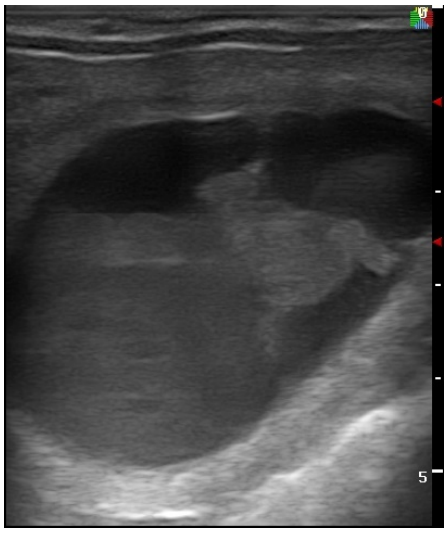

Figure 1. Ultrasound image of a fluid-filled mass with mixed echodensity, 35.7 x 44.7 mm, that extended from the posterior region of the uterine body into the cervical canal.

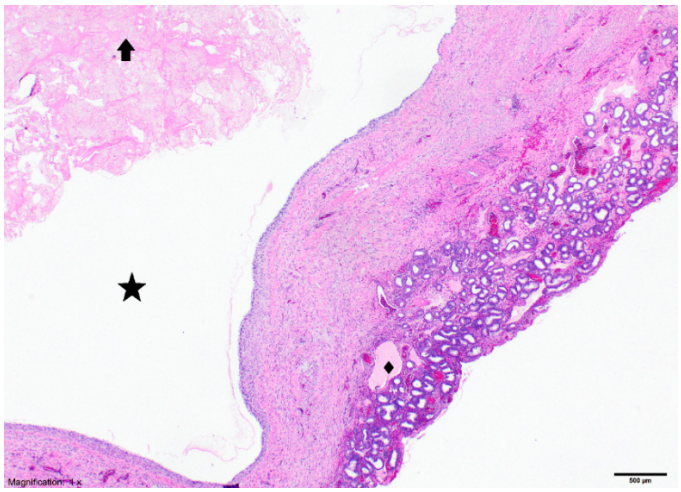

Figure 2. Histologic image of an endometrial lymphatic cyst and adjacent endometrial tissue stained with hematoxylin and eosin at 20x magnification. The cyst wall was lined by attenuated endothelium and the lumen (star) contained eosinophilic fibrillary material (arrow). The endometrium had dilated lymphatics (diamond), mild infiltrates of lymphocytes and plasma cells, as well as mild multifocal hemorrhage.