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Case Report

Schistosomus reflexus with another fetus in a beef heifer

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Abstract

Schistosomus reflexus (SR) is a rare, fatal fetal disorder observed in ruminants and other species. Defining features include spinal inversion, exposure of abdominal viscera, and limb abnormalities. Apparently, this is the first report documenting a case of SR with another fetus in a red Aberdeen-Angus beef heifer. Furthermore, a ventricular septal defect in a case of SR has not been documented before.

Keywords: Beef cattle, heifer, schistosomus reflexus, twins, cesarean surgery

Background

Schistosomus reflexus (SR) is a rare, fatal congenital disorder characterized by spinal inversion, exposure of the abdominal organs, and limb abnormalities in fetuses.[1] This condition has been reported in cattle, sheep, goats, buffalo, camels, rhinoceros, donkeys, cats, and nonmammalian species (e.g. sea turtles).[2–9] Abnormalities displayed in cases of SR suggested that the defect started early in embryos, possibly right after gastrulation, and possibly has a heritable component.[1]

SR is typically observed at parturition, most commonly presenting as dystocia. Abnormalities associated with the condition rendered vaginal delivery challenging or impossible, and often a cesarean surgery or fetotomy was necessary.[10] Because of poor prognosis in some cases, slaughter or euthanasia of the dam was considered. Multiple fetuses complicated handling SR cases. Multiple fetuses with 1 as SR and the other apparently normal were reported in dairy cattle, buffalo, sheep, and goats, but not in beef cattle.[11–14] In cattle, only 4.4% of twin fetuses were associated with a SR fetus.[10,11] Information on SR involving twins in beef cattle is limited. A beef cow presented for dystocia that had twins, with 1 displaying SR, is described below.

Case presentation

A 2-year red Aberdeen-Angus heifer (with ~ 6-hour history of dystocia) was presented. Parturition was on the expected day (based on the day of successful artificial insemination). Heifer was allowed to labor at owner's farm for several hours; owner palpated (transvaginal) the heifer and felt an abnormal calf and possibly viscera in the uterus. Primary veterinarian was called who referred the case.

Heifer was stable on presentation; however, her vital parameters were higher (rectal temperature: 40.2°C; pulse rate: 100 beats per minute, and respiratory rate: 50 breaths per minute), presumably from labor stress and ambient environmental conditions. Small intestinal loops were noticed protruding from her vulva and heifer had a bloody vulval discharge. Her ventral abdomen was covered with a mixture of mud and sweat, and she was excessively salivating. Fetal viscera were palpable (transrectally) within the vaginal canal and uterus.

Case management

A decision was made to perform cesarean surgery as the heifer had been in active labor for an extended period without progress. Fetotomy was not an option because of uncertainty (extent of fetal and possibly uterine pathology). Left paralumbar flank approach was adopted (heifer standing). An area behind the last rib to tuber coxae was clipped and prepped. After local anesthesia (inverted L block) with 2% lidocaine, abdomen was opened. An incision (~ 40 cm) was made and additional topical lidocaine blocks (with 2% lidocaine) were used on the musculature. The incision was 10 cm below the transverse processes of the lumbar vertebrae and 10 cm cranial to tuber coxae. Abdominal muscle layers were bluntly separated longitudinally from the muscle fiber orientation in a

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grid-like pattern. Peritoneum was then sharply incised for abdominal cavity entry.

Uterus was located and exteriorized. A forelimb of a fetus was palpated through the uterine wall and was used as a grip. Gentle traction was applied to bring the uterus to the abdominal incision. Approximately 30 cm full-thickness incision was made in the uterine wall parallel to palpated fetal limb. This allowed identification of fetal hindlimb. Obstetrical chains were placed on the hindlimbs and gentle traction was applied to the dead fetus. A second fetus was felt on further uterine palpation. Fetal hindlimbs were exteriorized and the fetus was removed from the uterus in a similar manner to first fetus. Gross abnormalities noted were consistent with SR. Uterus had 2 separate small, full-thickness lacerations (4 – 5 cm) dorsal and ventral to the uterine incision. Uterine tears and the primary incision were closed in 2 layers after the uterus was flushed copiously with sterile saline. First layer of each incision was closed (Utrecht pattern) using # 2 polyglactin 910 suture. Second layer was closed (Cushing inverted pattern) using # 2 polyglactin 910 suture and the uterus was placed back in the abdomen. Muscular layer was closed (simple continuous pattern) with # 3 polyglactin 910. Subcutaneous tissue was closed (simple continuous pattern) using # 2 polyglactin 910. Skin was closed (Ford interlocking pattern) with # 2 polymerized caprolactam. After closure, the incision was sprayed with an aluminum spray bandage and the heifer was given antiinflammatory (intravenous flunixin meglumine, 1.1 mg/kg) and perioperative antibiotic (intramuscular procaine penicillin G, 22,000 IU/kg). Heifer recovered uneventfully and walked to her stall unassisted.

Outcome

Calf 1 was a female red Aberdeen-Angus, appeared grossly normal, and had no outward abnormalities (identified prior to submitting for necropsy). Calf 2 was also a female red Aberdeen-Angus. This calf's appearance was consistent with SR and the small intestinal loops protruding from the heifer's vulva at presentation belonged to this calf. Calf appeared grossly normal, and the stillborn female calf was in good condition (body condition score [BCS] of 3 out of 5) and weighed 27.9 kg. Postmortem examination revealed abnormalities in heart and lungs, including hemorrhage in the right atrium, a small amount of red fluid in the pericardial sac. Abnormally dark and rubbery lungs indicated physiologic atelectasis. Findings confirmed that calf's death was due to dystocia (second fetus was lodged in the vagina preventing normal parturition).

Calf 2 was grossly abnormal (Figure 1); however, was in good condition (BCS 3 out of 5) and weighed 25.9 kg. Calf had prominent spinal curvature (lordosis) with failure of fusion of the body wall (noted along the thorax and abdomen). Due to failure of body wall closure, both the thoracic and abdominal viscera were exposed. All 4 limbs appeared abnormally angled with apparent fusion of the joints (ankylosis). Further examination of the viscera revealed a small, abnormal opening in the interventricular septum of the heart just below the aortic valve (referred as ventricular septal defect [Figure 2]). Diagnosis was SR (lordosis, exposure of thoracic and abdominal viscera, and limb).

Since a definitive cause of SR remained unknown, heifer samples were submitted for *Neospora caninum*, bovine respiratory syncytial virus, toxoplasmosis, infectious bovine rhinotracheitis, bovine viral diarrhea, and for selenium concentrations (Table 1).

Figure 1. Schistosomus reflexus calf (calf 2)

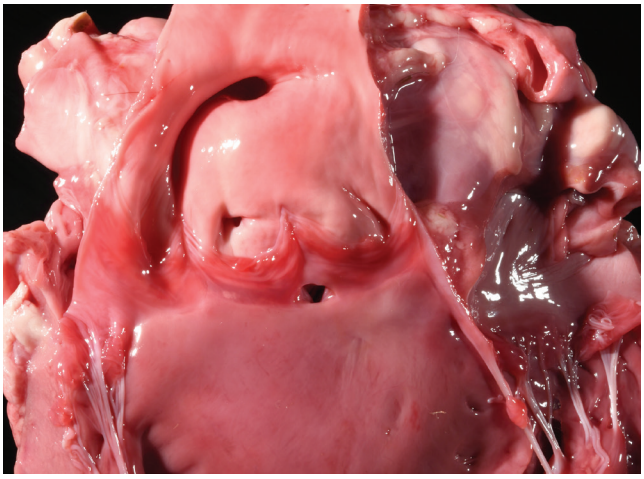

Figure 2. Ventricular septal defect (1 x 1 cm) noted in schistosomus reflexus calf (calf 2)

Table 1. Results of serological testing for the heifer

Sample	Test	Result
Serum	*Neospora caninum* – IFA	Negative
Serum	Bovine respiratory syncytial virus – IFA	Positive
Serum	Toxoplasmosis – IFA	Negative
Serum	Infectious bovine rhinotracheitis (IBR) Bovine viral diarrhea (BVD)	IBR 1:32; BVD 1:64
Serum	Selenium	0.06 ppm – marginal

After discharge, the cow was transitioned to oral meloxicam (0.5 mg/kg, once a day for 5 days) for pain control and intramuscular procaine penicillin G (22,000 IU/kg, twice a day for 3 days) for antibacterial coverage during initial recovery. Cow was allowed to rest (recovery from uterine surgery and healing of reproductive tract) for 4 – 5 months by her owner. After recovery interval, the cow was allowed to return to normal activity on pasture, where she was placed with a bull. After her first estrous cycle, she was examined

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Table 2. Cases of twin ruminants with SR

Species	Findings	Twin outcome
Bovine (Holstein cross)[22]	Exposed abdominal viscera, marked retroflexion of spine, anterior and lateral positioning of the pelvic bones, sacrum, and hind limbs	Normal
Caprine[14]	Mummified fetus, marked ventral spinal curvature, lateral bending of fetal body and chest wall, incomplete closure of abdominal wall with exposed viscera	Stillborn
Water buffalo[21]	Thoracic and abdominal viscera exposed, spinal inversion, enlarged liver and rumen	Conjoined
Caprine[16]	Ventral spinal curvature, exposed abdominal viscera, incomplete closure of abdominal wall	Conjoined
Ovine[15]	Thoracic and abdominal viscera exposed, mummified fetuses, ventral spinal curvature	Conjoined
Bovine (Holstein)[11]	Prognathism, wry nose, lordosis, atresia ani, bifurcated scrotum, dorsally deviated sternum, total eventration of abdominal and thoracic viscera	Freemartin
Ovine[13]	Kyphoscoliosis of thoracic vertebrae, exposed abdominal viscera due to failure of abdominal wall closure	Conjoined
Ovine[23]	Thoracic and abdominal viscera exposed, incomplete closure of chest wall, lateral reflection and ankylosis in lumbar and thoracic regions	Normal
Caprine[24]	Thoracic and abdominal viscera exposed, acute angulation and ankylosis of lumbar vertebrae	Normal

(transrectal palpation) for pregnancy and was determined to be pregnant. Cow's pregnancy progressed normally for the first 4 – 5 months and was sold to another beef cattle producer. At term, the cow delivered a normal calf; further reproductive information is not available (inability to contact the owner).

Discussion

In bovine practice, SR is not a common occurrence, with limited numbers suggesting that it accounted for < 1% of dystocias in certain cattle populations.[10] Cases of SR involving twins or multiple fetuses are more limited, with some examples in dairy cattle,[1,11] lambs,[13,15] kids,[14,16,17] buffalo,[18] camels,[5] cats,[19] dogs,[19] and foals.[20] Although SR has occurred in ruminants with twins (Table 2), this is the first report documenting a case of SR in a beef cattle heifer with twins and a ventricular septal defect (VSD).

Defining features of SR in ruminants include spinal inversion, exposure of abdominal viscera, limb ankylosis, positioning of the legs adjacent to the skull, and lung and diaphragm hypoplasia.[1] In this case, exposure of abdominal viscera and severe spinal inversion and a membranous (VSD) were present. Latter defect was not a common finding in other ruminant SR cases.[1] Although not previously reported in conjunction with SR cases, VSD remains the most common congenital cardiac defect in ruminants, commonly, in the region below the aortic valve (as observed in this case) in cattle.[25,26] Prevalence of SR in cattle has been reported (0.01[27] – 1.3%[10]), encompassing both dairy (80%) and beef cattle breeds. To authors' knowledge no such prevalence report exists for SR in Red Angus cattle.

Dystocia requiring intervention is a common occurrence with SR cases in bovine practice.[28] Intervention selection is largely dependent on presentation and position of the fetus,[21] and the duration of stage 1 of parturition. Factors (e.g. fetal size in relation to vaginal canal) and the expertise of available clinicians cannot be overlooked.[29] SR prevalence rate was 0.93% (16 cases out of 1,716 dystocias).[27] Twin births had a higher incidence of dystocia due to malpresentation of 1 fetus or simultaneous presentation of both fetuses.[30] Most common methods of correcting dystocias include manual manipulation and delivery of fetus, fetotomy, or via cesarean surgery. Cesarean surgery was conducted in 25.6% of cases, fetotomy was performed in 56.7%, and manual traction was successful in only 3.3% of cases.[10] Mild traction and manual manipulation of the fetus, whether by use of obstetrical chains or other means, is generally pursued first and is a less invasive option.[10,31] This method of manipulation was more often reported successful in small ruminant cases of SR than in cattle, unless the SR fetus was smaller than a normal full-term calf.[4] If manual manipulation fails, the secondary method is dependent on whether the fetus is viable and whether sufficient space in the uterus and vaginal canal is available for introduction and use of a fetotome. If fetotomy is an option based on space and the fetus has been confirmed dead, a partial or full fetotomy can be performed. In cases of SR, fetotomies are commonly performed when the degree of spinal curvature prevents passage of fetus through the birth canal.[21] Cesarean surgeries are frequently performed due to inadequate space in the uterus or birth canal and the inability to otherwise reposition the fetus or manipulate a fetotome safely.[9,21] Surgical management of dystocia allows for a more thorough examination of the reproductive tract of the cow and is the method of choice if presentation of the dystocia was delayed from onset of parturition.[32] In each method described,

medical management (e.g. caudal epidural) and providing adequate sedation and pain control should be executed.

Although currently it is believed that SR is heritable condition in ruminants,[1] recent work in sea turtle has suggested a toxicity or epigenetic etiology.[33] In olive ridley sea turtle (*Lepidochelys olivacea*) embryos with SR malformations and without malformations, a positive correlation was identified with mercury concentrations and DNA methylation in embryos with SR abnormalities.[22] Multiple drugs and compounds caused changes in DNA methylation and presented with epigenetic or quasiepigenetic responses, including beta-lactam antibiotics, lithium, and opioids.[34] Currently, the association is between SR in cattle and any toxicities or epigenetic changes is unknown. Future studies should investigate the role of either of these potential influences on SR in ruminants.

Learning points

- SR is a rare and fatal condition observed in cattle. Defining features of the condition include spinal inversion, exposure of abdominal viscera of fetus.
- A case of SR (with ventricular septal defect) with a normal twin fetus in a Red Angus beef heifer is documented.
- Although fetotomies are the most frequently utilized method of extracting a single SR fetus, in cases of twins, cesarean surgery may be necessary due to inadequate space (uterus and birth canal) for manipulation.

Conflict of interest

None to declare.

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