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Case Report

Congenital hydrocephalus in a stillborn Haflinger foal

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Abstract

A 14-year, multiparous Haflinger mare, apparently fullterm (unknown breeding or ovulation date), was presented for dystocia. Mare was transvaginally palpated at the farm by the referring veterinarian; foal was in craniolongitudinal presentation, dorsosacral position with extended forelimbs in the vaginal vault and head just cranial to cervix, and a cranial abnormality of the foal prevented assisted vaginal delivery. Mare was referred. Foal was not alive at presentation and was undeliverable with assistance because of congenital hydrocephalus. Anesthetized mare was placed in Trendelenburg position, controlled vaginal delivery was elected and foal was removed via fetotomy. Mare was negative for β1, 3- *N*-acetylgalactosaminyltransferase 2 (B3GALNT2) nonsense mutation (reported to be responsible for hydrocephalus). Dystocia in a Haflinger mare because of hydrocephalus is reported for the first time.

Keywords: Mare, Haflinger fetus, congenital hydrocephalus, B3GALNT2 gene

Introduction

Congenital hydrocephalus (CH) is an inborn abnormality where cerebrospinal fluid (CSF) accumulates and dilates brain ventricles.[1–5] Congenital hydrocephalus can be internal, external, communicating, or noncommunicating depending on the location of CSF accumulation in brain. Dilation occurs either from increased production or decreased absorption of CSF. In internal hydrocephalus, CSF accumulates within ventricles whereas in external hydrocephalus, CSF accumulates outside ventricles. Communicating hydrocephalus is defined as a problem with absorption, and noncommunicating hydrocephalus occurs when there is an obstruction to absorption of CSF.[2,4,6] Congenital hydrocephalus has been noted in multiple species including humans,[7] rats,[8] rabbits, ruminants, birds, cats, dogs,[6] and many horse breeds (Friesian, Standardbred, Belgian, Quarter horse, and Miniature horse).[9]

In horses, hydrocephalus is still a rare occurrence with an incidence of 0.6 per 1,000 births.[6] Equine CH has been studied in the Friesian population where the average inbreeding coefficient is higher. For this breed, there is a commercial test of the nonsense mutation of β1, 3- *N*-acetylgalactosaminyltransferase 2 (B3GALNT2)[1,2,4] that identifies carriers of the hydrocephalus inducing autosomal recessive gene.[1,2,4] Mutation of B3GALNT2 gene has also been detected in a Belgian Draft horse.[4] In humans, only 40% of hydrocephalus cases are believed to be genetically related to an X-linked recessive trait[7,10] Other known causes of hydrocephalus in other species (dogs, cats, cattle, and pigs) include viral and bacterial causes.[6] There are 2 reports in the same publication of 2 aborted Halfinger foals that had hydranencephaly. These foals were sired by the same stallion.[10] To date, there is no published case report of a presumed fullterm hydrocephalus foal in a Haflinger mare.

Case presentation

Case history

A 14-year, multiparous Haflinger mare, apparently fullterm (unknown breeding or ovulation date), was presented for dystocia. Mare had 6 foals before and last foaling was a normal delivery. Mare was transvaginally palpated at the farm by the referring veterinarian; foal was in craniolongitudinal in presentation, dorsosacral in position with extended forelimbs in the vaginal vault and head just cranial to cervix, and a cranial abnormality of the foal prevented assisted vaginal delivery. Mare was referred.

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Clinical examination

At presentation, mare had been in Stage 2 of parturition for ~ 6 hours. Initial examination revealed that the mare was tachycardic, tachypneic, had a small amount of dried blood on the right nostril, and had a mild amount of hemorrhagic discharge from the vulva. Mare's body condition was scored as 7/9. Total protein was 6.4 g/dl, PCV was 38%, and lactate was 1.6 mmol/l. Transabdominal ultrasonography was performed during left jugular vein catheterization; fetus was confirmed dead (visualization of lack of heartbeat). Mare was given 350 mg of intravenous xylazine. Tail was wrapped, and vulva was washed with a nonresidue soap. Two minutes later, mare was given 1,200 mg of intravenous ketamine and 200 mg of intravenous propofol for induction of anesthesia. An endotracheal tube (26 mm) was placed, and mare was maintained on sevoflurane gas mixed with 100% oxygen, with lidocaine and dobutamine as a constant rate infusion. After being initially placed in the Trendelenburg position, where the mare was in dorsal recumbency with the hind legs elevated via hoist, mare's blood pressure increased which indicated that anesthetic plane became light, so a bolus of 30 mg of intravenous butorphanol was given over 10 minutes. Fetus was transrectally palpated and was in craniolongitudinal presentation, dorsosacral position with extended forelimbs in the vaginal vault. Foal's head was extended but positioned just cranial to the pelvic brim, and an irregularly-shaped cranium was palpated. Cesarean surgery was offered and declined.

Treatment

Due to enlarged cranium and the inability to perform cesarean surgery, a partial fetotomy was elected to extract the foal. First cut was made transversely through the enlarged cranium, and the second was made between C1 and C2 vertebrae (Figures 1 and 2). Fetal skull had an incomplete boney cranium with the bone forming around the lateral aspects and a 1-inch strip of bone forming down the sagittal midline on the dorsal aspect (Figure 3). After the cranial cuts were made, resultant fragments were removed. Chains were placed on the forelimbs with a loop above fetlock and half hitch below fetlock joints (chain running dorsally on the forelimbs) and foal was delivered with manual traction. Mare's nongravid uterine horn intussuscepted during delivery and was promptly replaced. Fetal membranes remained tightly adhered to the endometrium. Mare recovered from anesthesia and tetanus toxoid vaccine was given.

Outcome

Foal had a cranium volume of 3,721 cm^3 that equates to ~ 1 gallon (3.71 liters). Dimensions of foal's cranium were as follows: 20.95 cm (rostral to caudal), 18.4 cm (width), and 18.4 cm (dorsal to ventral) (Figure 4). Foal's brain was ~ 3 mm in thickness (Figure 5). Primary differential diagnosis was an internal congenital hydrocephalus. Mare was routinely treated for retained fetal membranes with a combination of antibiotics, antiinflammatory agents, ecbolics, and uterine lavage. Uterine lavage was performed twice daily with 5-10 liters of lactated Ringer's Solution. Intramuscular oxytocin (5 IU) was given every 2 hours until fetal membranes were expelled. Antibiotics (intravenous potassium penicillin 22,000 IU/kg every 6 hours and intravenous gentamicin 6.6 mg/kg once daily) and an antiinflammatory (intravenous flunixin meglumine 1.1 mg/kg every 12 hours). Mare shed fetal membranes 3 days after dystocia and was discharged from the hospital without any further complication. Owners elected not to breed the mare during the subsequent season.

Figure 1. Location of fetotomy first cut.

Genetic testing was authorized by the owner. Since B3GALNT2 mutation has previously been reported in a Belgian mare and there are no other genetic variants that have currently been identified, 20-30 hairs with roots were submitted to UC Davis Veterinary Genetics Laboratory for analysis. This gene is autosomal recessive, and the stallion was not available for testing. Final results indicated that the mare did not carry the recessive trait for this mutation.

Discussion

Congenital hydrocephalus is reported to affect 0.6 out of 1,000 births in horses.[6] Currently, there is only 1 commercial test available for Friesian mares with the identified nonsense mutation of B3GALNT2.[1,2,4] Congenital hydrocephalus is present in multiple species, with 40% of human cases stemming from a genetic cause.[7] In ruminants, hydrocephalus is often associated with teratogenic viruses, whereas in felids and birds there are reports associating hydrocephalus with vitamin A deficiency. Additionally in dogs, CH has been linked to anatomic defects of the skull, vertebral growth, and craniovertebral junction.[6] In contrast, there have been no reports of vitamin deficiency or

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viral causes of hydrocephalus in horses and the ultimate cause is not well described in the literature.[8]

To authors' knowledge, this is the first case report of a Haflinger mare with a hydrocephalus foal at term. A Belgian draft horse had a hydrocephalic foal that was positive for B3GALNT2 mutation.[4] In our case, the Haflinger mare did not carry the B3GALNT2 mutation, suggesting that there is either another genetic mutation or another cause of hydrocephalus that has not yet been identified. There are no reported cases in horses of viral illness causing hydrocephalus.[6] Further research is needed to provide insight into different genetic and nongenetic causes of hydrocephalus in horses and provide broader diagnostic tests and possible preventions for this fatal condition.

Figure 2. Location of fetotomy second cut.

Learning points

- Congenital hydrocephalus in horses is rare; however, in certain breeds can be associated with a mutation in B3GALNT2 gene.
- Not all cases of fetal hydrocephalus are caused by a mutation in B3GALNT2 gene; apparently, there are other causes.
- Hydrocephalic fetuses can complete pregnancy; however, will likely result in dystocia.

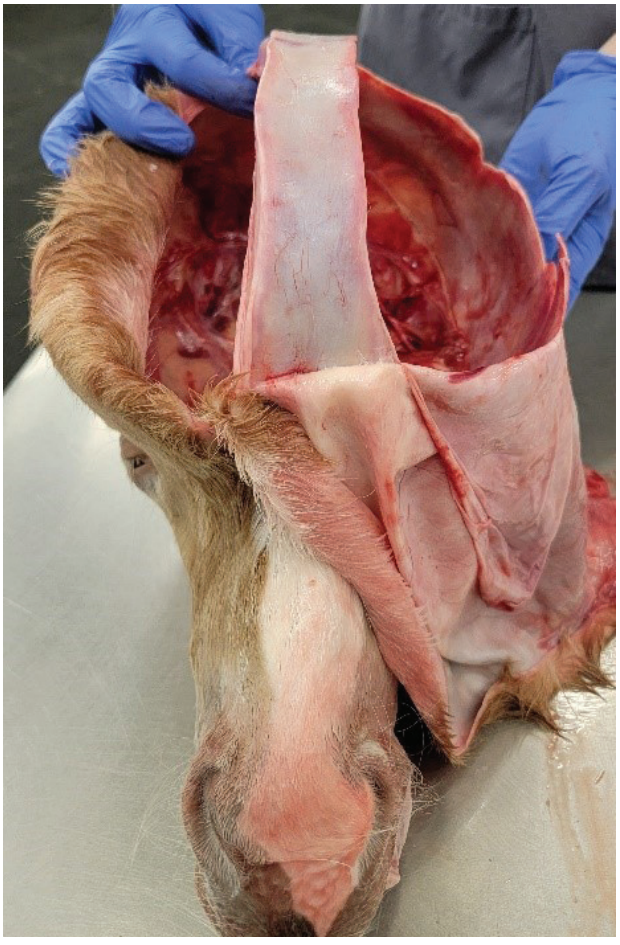

Figure 3. Lateral formation of cranium with a boney strap down the sagittal midline.

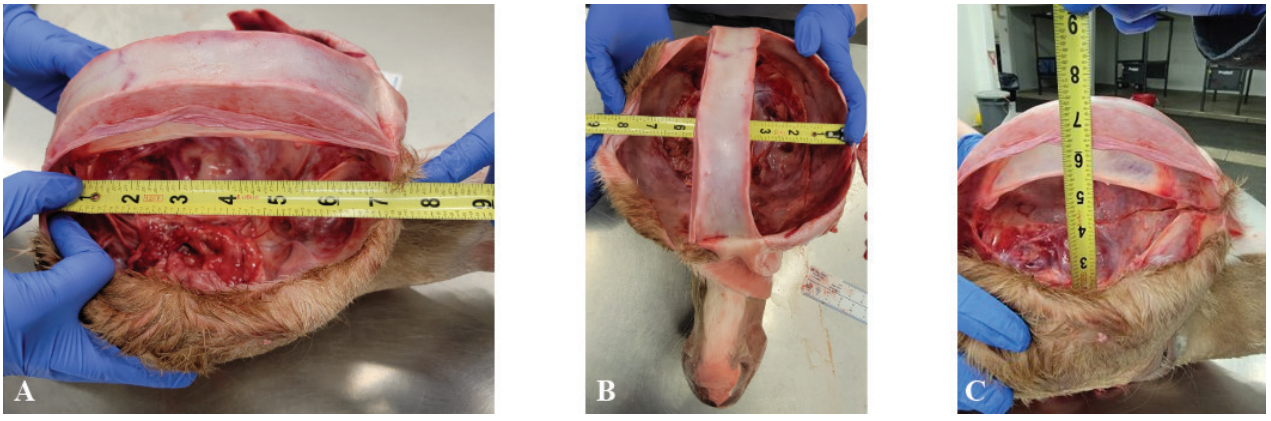

Figure 4. Dimensions of foal's cranium; A. rostral to caudal, B. width, C. dorsal to ventral.

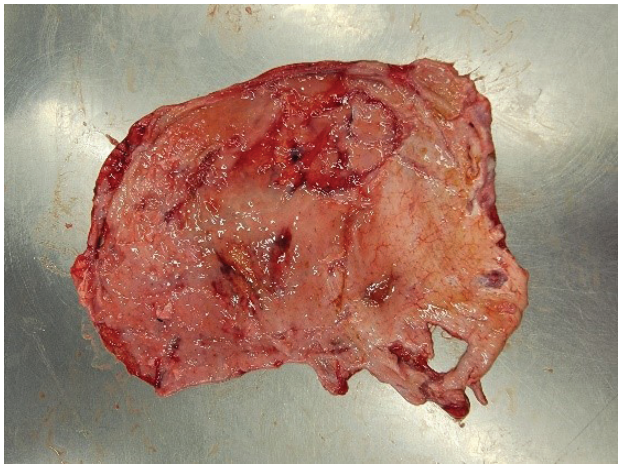

Figure 5. Brain matter from foal's cranium.

Conflict of interest

None to Report.

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