Reduction Glossoplasty in a Calf with Bifid Tongue

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ABSTRACT

Background: Fetal malformations are characterized by anatomical changes that compromise an organ or system. Tongue formation in bovines occurs with the fusion of three structures at the end of the fourth week of gestation, and any failure during this stage of embryonic development may lead to tongue malformation. Bifid tongue, also called glossoschisis, is a rare congenital abnormality in any species and is characterized by incomplete fusion of the lateral tongue buds, resulting in a deep groove in the midline of the tongue. The objective of this study was to describe a case of bifid tongue and the procedure of reduction glossoplasty in a calf of the Girolando breed in Tocantins State.

Case: A male mixed-breed (Holstein-Friesian × Gir) calf, born from natural mating in the municipality of Araguaína, Tocantins, was clinically assessed in the Sector of Ruminant Clinical Medicine of the Federal University of Tocantins at 2 months of age. The owner reported that the animal exhibited difficulty in suckling after birth and that on inspection of the oral cavity, he observed changes in the tongue and mandible. Clinical examination of the oral cavity revealed the presence of a bifid tongue and abnormal fusion of the mandible in the region of the lower incisive teeth. It was decided to perform a surgical procedure with the aim of improving the animal's quality of life because the owner wanted to keep it in the farm. An incision was made, followed by removal of the medial rims of the two tines of the tongue, and synthesis was performed, joining the ventral rims of the tongue, beginning at the root and ending at the apex.

Discussion: The observed lesions are compatible with bifid tongue and mandibular fissure, both previously reported in other species. The literature has no reports of bifid tongue in bovines. The etiology of fetal malformations is still unclear, and the primary causes known in Brazil include ingestion of toxic plants by the mother, such as *Mimosa tenuiflora* and *Poincianella pyramidalis*; infections, with the main viral agents being bovine viral diarrhea virus and blue tongue virus; and teratogenic agents, namely certain medications administered during embryogenesis, e.g., ivermectin. Other potentially teratogenic agents that have not been identified as causes of malformation include radiation, cortisone, benzimidazoles, sulfonamides, folate antagonists, and organophosphates. The intense genetic improvement that the Holstein-Friesian breed has been subjected to, including inbreeding that results in consanguinity, may be a determining factor for the breed carrying mutant alleles. Because the calf in this report was crossbred from parents with Holstein-Friesian ancestry, it could carry mutant alleles that led to the malformation. Another etiology proposed in a study on embryological bases by Goodacre and Wallace (1990) is the persistence of buccopharyngeal membrane and amniotic constriction bands in the region of the branchial arches; this cause cannot be ruled out in the present case. Calves born with fetal malformations generate losses for cattle breeders because the calf dies either before or after birth, which may culminate with the death of the mother or, in cases in which the anomaly is compatible with life, damage to the animal's development and well-being, as in the present report. Bifid tongue is a rare malformation in bovines, and reduction glossoplasty is essential for the description and improvement of techniques that aid bovine medicine; however, animals subjected to this procedure should not be used for reproduction.

Keywords: congenital malformation, bifid tongue, surgical correction.

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INTRODUCTION

Fetal malformations are characterized by anatomical changes that compromise an organ or system. They may occur in the embryonic or fetal stages and lead to miscarriage or neonatal death, in addition to reproductive damage [5,12,13,19]. Several causes may be involved in the pathogenesis of malformations, such as physical factors; infections caused by viruses, bacteria, and protozoa; mineral deficiencies; intoxication by plants; genetic inheritance; drugs; and radiation [12].

Tongue formation in bovines occurs with the fusion of three structures at the end of the fourth week of gestation. These structures fuse to form the free part of the tongue, which is capable of moving inside the oral cavity. Any failure during this stage of embryonic development may lead to malformation of the tongue [7,8].

Bifid tongue or glossoschisis is a rare congenital abnormality in any species [25] and is characterized by the incomplete fusion of the lateral tongue buds, thus resulting in a deep groove in the midline of the tongue [8]. It has been previously described in mules, in which it is occasionally associated with lesions in mandibular fusion [25], dogs [27], bovines [4], and humans [11,26].

The diagnosis of congenital malformations is challenging for veterinarians. Because investigation of the potential causes of fetal malformations is typically performed only when a certain defect occurs frequently within the same herd or geographical area, few malformations are reported [17,18].

The objective of this study was to describe a case of bifid tongue and the procedure of reduction

glossoplasty in a calf of the Girolando breed in Tocantins State.

CASE

A mixed-breed (Holstein-Friesian × Gir) calf born from natural mating in the municipality of Araguaína, State of Tocantins, Brazil, was clinically assessed at 2 months of age in the Sector of Ruminant Clinical Medicine (SCMR) of the Federal University of Tocantins. The owner reported that the animal exhibited difficulty in suckling after birth, and on inspection of the oral cavity, he observed changes in the tongue and mandible. Therefore, artificial feeding was initiated, and the animal was kept under observation in the farm (Figure 1).

Clinical examination of the oral cavity revealed the presence of bifid tongue and abnormal fusion of the mandible in the region of the lower incisive teeth. The forked tongue was split into two tines, from the root to the apex, and each tine measured 18 cm (Figure 2).

At the time of the clinical assessment, the calf had a good body score and did not exhibit other visible congenital malformations. The surgical procedure was performed with the aim of improving the animal's quality of life because the owner wanted to keep it in the farm (Figure 3).

The surgical procedure was performed in the farm where the animal was kept. The calf was sedated with a combination of xylazine hydrochloride (Rompun 2%® - 0.4 mg/kg)¹ and acepromazine (Acepran 1%® - 0.6 mg/kg)² via intramuscular (IM) administration, according to the method described [21]. Infiltration local anesthesia was administered in the perimeter of the tongue tines using lidocaine 2% with a vasoconstrictor.

Figure 1. Girolando calf presenting with bifid tongue and changes in the fusion of the mandibula.

Figure 2. Cleft of the tongue from root to apex.

Figure 3. Preoperative demarcation before glossoplasty.

Figure 4. Aspect immediately after surgical correction.

After sedation and local anesthesia, asepsis of the tongue and oral cavity with povidone-iodine 3% was performed. An incision was made, and the medial rims of the two tongue tines were excised. Subsequently, synthesis was performed, joining the ventral rims of the tongue from the root to the apex of the tongue. They were then joined with simple interrupted suture on the dorsal surface of the tongue using nylon thread N°0, thus completing the reduction glossoplasty procedure that reshaped the calf's tongue (Figure 4). Postoperatively, the animal received IM enrofloxacin (Kinetomax® - 2.5 mg/kg)¹ once a day for 5 days and IM flunixin meglumine (Banamine® - 1.1 mg/kg)³ once a day for 3 days. Following surgery and recovery from sedation, the calf was placed with its mother and was able to suckle.

DISCUSSION

The observed lesions are compatible with bifid tongue and mandibular fissure, both previously reported in other species [10,25,27].

The literature has no reports of bifid tongue in bovines. That most congenital defects are rare and some are so rare that the opportunities to observe them are very few [9]. Therefore, description of cases of fetal malformations in bovines is important in order to create an inventory of cases and aid in the investigation of the potential causes.

The etiology of fetal malformations is still unclear, and the main causes known in Brazil include the ingestion of toxic plants by the mother, such as *Mimosa tenuiflora* [5,23] and *Poincianella pyramidalis* [4,20,24], but these plants were not seen during an inspection of the property as they are native to the semi-arid region of Northeastern Brazil, a different

region; and infections, with the main viral agents being the bovine viral diarrhea virus (BVDV) [6,22] and blue tongue virus [2,14,15]. It was not possible to perform serology tests of the calf and its mother; therefore, infection by BVDV cannot be ruled out as the cause of the malformation. Some teratogenic agents, namely certain medications given during embryogenesis [16], e.g., ivermectin [3], have also been postulated as causes of fetal malformations. The breeder reported the use of doramectin to control endo- and ectoparasites; however, a change of medication and administration of ivermectin to the mother during embryogenesis is possible, and it is therefore not possible to rule out its use as the cause of the malformation. Other potentially teratogenic agents that have not been positively identified as causes of malformation include radiation, cortisone, benzimidazoles, sulfonamides, folate antagonists, and organophosphates [16]. Genetic causes are frequently reported in cases of fetal malformations in bovines [28]. The intense genetic improvement that the Holstein-Friesian breed has been subjected to, including inbreeding that results in consanguinity, may be a determining factor for the breed carrying mutant alleles [1]. Because the calf in this report was a crossbreed from parents with Holstein-Friesian ancestry, it could carry mutant alleles that led to the malformation. Another etiology proposed in a study on embryological basis by Goodacre and Wallace [9] is the persistence of the buccopharyngeal membrane and amniotic constriction bands in the region of the branchial arches; this cause cannot be ruled out in this case of malformation.

Calves born with fetal malformations generate losses for cattle breeders because the calf dies either

before or after birth, which may culminate with the death of the mother or, in cases in which the anomaly is compatible with life, damage to the animal's development and well-being, as in the present report. It is necessary that upon identifying a fetal malformation that is compatible with life, the veterinarian explains to the cattle breeder that the affected animal should not be used for reproduction because there is a risk of disseminating the genes of the fetal malformation among the herd. It was not possible to identify the causative agent of this calf's fetal malformation (bifid tongue) in the present report, but the problems involved in the animal's development and its general condition were explained to the cattle breeder. It is necessary to continue investigating the potential causes of fetal malformations in ruminants across Brazil.

Surgical correction was selected because the breeder wanted to keep the animal alive and provide it with a better quality of life; however, he was informed that the animal's weight gain could be compromised and that reproduction was not recommended. Thus, orchiectomy was indicated as soon the animal recovered from the first surgical procedure. Rifai *et al.* [25] reported that three mules were destined for traction following reduction glossoplasty and correction of mandibular fissure.

The forked tongue prevented the calf from naturally suckling its mother's milk. Therefore, it was necessary to initiate artificial feeding to keep the animal alive and with a good body score until reduction glossoplasty was performed. If the owner had not opted for surgical correction, the animal's quality of life would have deteriorated because it would need to start eating solid foods (grass), which would be difficult to bite, chew, swallow, and ruminate.

Rifai *et al.* [25] reported the case of a 12-day-old mule with a history of not being able to suckle that was referred to a society for the protection of animals in Morocco. Assessment of the buccal cavity revealed the presence of a bifid tongue and mandibular fissure, and the animal was referred for correction surgery and postoperative treatment. After 5 days, the mule's

clinical parameters were affected and it died. The findings described by are similar to those observed in the present study, i.e., the animal was unable to suckle. However, this case was different because the calf was artificially fed by the breeder before the surgical procedure. That kept it in a good physical condition and it was able to suckle from the cow postoperatively. The mandibular fissure was not corrected because the procedure is invasive and there was a risk of causing more postoperative complications to the animal, especially as the procedure was performed in the property.

Performed mandibular corrections in three mules that exhibited mandibular bone fusion failure and mucosa overlap. The surgical procedure consisted of uncovering the medial extremities of each half mandible, which were curetted and fixated with copper wire around the incisive teeth to stabilize the symphysis. The procedure was unsuccessful, the vital parameters of two animals were compromised, and the mules died; the other mule, which also had ankyloglossia, recovered from the surgical procedure and resumed suckling [25], as was the case in the present report.

There is little data in the literature regarding the surgical procedures used in the correction of fetal malformations because a number of cattle breeders prefer to euthanize the animal immediately after the malformation is identified. Moreover, veterinarians do not perform the surgery because there is a chance of propagating the mutant genes of these animals in the future.

Bifid tongue is a rare malformation in bovines, and performing reduction glossoplasty is important for the description and improvement of techniques that aid bovine medicine; however, animals subjected to the procedure should not be used for reproduction.

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REFERENCES

- 1 Agerholm J.S., Bendixen C., Andersen O. & Arnbjerg J. 2001. Complex vertebral malformation in Holstein calves. *Journal of Veterinary Diagnostic Investigation*. 13(4): 283-289.
- 2 Antoniassi N.A.B., Pavarini S.P., Ribeiro L.A.O., Silva M.S., Flores E.F. & Driemeier D. 2010. Alterações clínicas e patológicas em ovinos infectados naturalmente pelo vírus da língua azul no Rio Grande do Sul. *Pesquisa Veterinária Brasileira*. 30(12): 1010-1016.

- 3 Ayres M.C.C. & De Almeida M.A.O. 1999. Agentes antinematódeos. In: Spinosa H. S., Gorniak S.L. & Bernardi M.M. (Eds). *Farmacologia Aplicada à Veterinária*. 2.ed. Rio de Janeiro: Guanabara Koogan, pp.460-463.
- 4 Correia D.A.D.B., Neto M., Gomes D.L.D.S. & Torres M.B.A.D.M. 2017. Malformações congênicas e abortos induzidos experimentalmente pela ingestão de *Poincianella pyramidalis* (Tul.) LP Queiroz (catingueira) em ovelhas. *Pesquisa Veterinária Brasileira*. 37(12): 1430-1436.
- 5 Dantas A.F.M., Riet-Correa F., Medeiros R.M.T., Galiza G.J.N., Pimentel L.A., Anjos B.L. & Mota R.A. 2010. Malformações congênicas em ruminantes no semiárido do Nordeste Brasileiro. *Pesquisa Veterinária Brasileira*. 30(10): 807-815.
- 6 Dezen S., Otonel R.A.A., Alfieri A.F., Lunardi M. & Alfieri A.A. 2013. Perfil da infecção pelo vírus da diarreia viral bovina (BVDV) em um rebanho bovino leiteiro de alta produção e com programa de vacinação contra o BVDV. *Pesquisa Veterinária Brasileira*. 33(2): 141-147.
- 7 Emmanouil-Nikoloussi E.N. & Kerameos-Foroglou C. 1992. Developmental malformations of human tongue and associated syndromes (review). *Bulletin du Groupement international pour la recherche scientifique en stomatologie & odontologie*. 35(1-2): 5-12.
- 8 Garcia S.M.L. & Fernández C. G. 2001. *Embriologia*. 2.ed. Porto Alegre: Artmed, 294 p.
- 9 Goodacre T.E.E. & Wallace A.F. 1990. Congenital alveolar fusion. *British Journal of Plastic Surgery*. 43 (8): 203-209
- 10 Hiebert J.C., Johnson A.B., Tran H.H., Yu Z. & Glade R.S. 2016. Congenital tongue mass with concomitant cleft palate and bifid tongue: a case report and review of the literature. *The Cleft Palate-Craniofacial Journal*. 53(2): 245-248.
- 11 James A.W., Culver K., Hall B. & Golabi M. 2007. Bifid tongue: a rare feature associated with infants of diabetic mother syndrome. *American Journal of Medical Genetics Part A*. 143(17): 2035-2039.
- 12 Jolly R.D. 2002. Screening for genetic diseases in cattle. *Australian Veterinary Journal*. 80(5): 284-285.
- 13 Khodakaram-tafti A. & Ikede B.O. 2005. A retrospective study of sporadic bovine abortions, stillbirths, and neonatal abnormalities in Atlantic Canada, from 1990 to 2001. *Canadian Veterinary Journal*. 46(7): 635-637.
- 14 Lager I.A. 2004. Bluetongue virus in South America overview of viruses, vectors, surveillance and unique features. *Veterinaria Italiana*. 40(3): 89-93.
- 15 Leipold H.W. & Dennis S.M. 1986. Congenital defects affecting bovine reproduction. In: Morrow D.A. (Ed). *Current therapy in theriogenology: diagnosis, treatment and prevention of reproductive diseases in small and large animals*. Philadelphia: W.B. Saunders Company, pp.177-199.
- 16 Leipold H.W., Dennis S.M. & Huston K. 1972. Congenital defects of cattle: Nature, cause, and effect. *Advances in Veterinary Science and Comparative Medicine*. 16: 103-150.
- 17 Leipold H.W., Huston K. & Dennis S.M. 1983. Bovine congenital defects. *Advances in Veterinary Science and comparative medicine*. 27: 197-271.
- 18 Macedo J.T.S.A., Lucena R.B., Giaretta P.R., Kommers G.D., Figuera R.A., Irigoyen L.F. & Barros C.S.L. 2011. Defeitos congênicos em bovinos da Região Central do Rio Grande do Sul. *Pesquisa Veterinária Brasileira*. 31(4): 297-306.
- 19 Marcelino S.A.C., Macêdo J.T.S.A., Reis S.D.S., Lacerda M.S.C., Silva A.R.S., Riet-Correa F., Pimentel L.A. & Pedroso P.M.O. 2017. Malformações em pequenos ruminantes no semiárido da Bahia: aspectos epidemiológicos, clínico-patológicos e radiológicos. *Pesquisa Veterinária Brasileira*. 37(12): 1437-1442.
- 20 Moore K. & Persaud T.V.N. 2000. Defeitos congênicos humanos. In: *Embriologia Clínica*. Rio de Janeiro: Guanabara Koogan, pp.161-189.
- 21 Muir III W.W., Hubbell J.A.E., Skarda R.T. & Bednarski R.M. 2001. Fármacos usados na medicação pré-anestésica. In: *Manual de Anestesia Veterinária*. Porto Alegre: Artmed, pp.31-44.
- 22 Pavarini S.P., Sonne L., Antoniassi N.A.B., Santos A.S., Pescador C.A., Goberllini L.G. & Driemeier D. 2008. Anomalias congênicas em fetos bovinos abortados no sul do Brasil. *Pesquisa Veterinária Brasileira*. 28(3):149-154.
- 23 Pimentel L.A., Correa F.R., Gardner D., Panter K.E., Dantas A.F.M., Medeiros R. M.T. & Araújo J.A.S. 2007. *Mimosa tenuiflora* as a cause of malformations in ruminants in the northeastern Brazilian semiarid rangelands. *Veterinary Pathology*. 44(6): 928-931.

- 24 Reis S.D.S., Oliveira R.S., Marcelino S.A.C., Macêdo J.T.S.A., Riet-Correa F., Pimentel L.A. & Pedroso P.M.O. 2016.** Congenital malformations and other reproductive losses in goats due to poisoning by *Poincianella pyramidalis* (Tul.) L.P. Queiroz (*Caesalpinia pyramidalis* Tul.). *Toxicon*. 118: 91-94.
- 25 Rifai S., Bouayad H., Kay G., Knottenbelt D.C. & Smith M. 2006.** Bifid tongue and mandibular cleft in three mule foals. *Veterinary Record*. 158(3): 97.
- 26 Surej K.L., Kurien N.M. & Sivan M.P. 2010.** Isolated congenital bifid tongue. *National journal of maxillofacial surgery*. 1(2): 187.
- 27 Villagomez D.A. & Alonso R.A. 1998.** A distinct Mendelian autosomal recessive syndrome involving the association of anotia, palate agenesis, bifid tongue, and polydactyly in the dog. *Canadian Veterinary Journal*. 39(10): 642.
- 28 Whitlock B.K., Kaiser L. & Maxwell H.S. 2008.** Heritable bovine fetal abnormalities. *Theriogenology*. 70(3): 535-549.