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Bifid tongue and mandibular cleft in a foal: a case report

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Abstract

The aim of this report is to describe a succesfull surgery intervention to correct a bifid tongue and mandibular cleft in a brazilian foal. Despite many references showed unsuccesfull outcome of few cases, the present one describes a simple but intensive follow up that resulted in a good prognosis and a better quality of life to the animal. Reductive glossoplasty, two screws transecting the hemimandibles and cerclage through incisors was performed to stabilize the separated tissues. Antibiotic and antinflamatory therapies was conducted in the post operatory period and the ability to suckle and eat was imperative to the success of the outcome.

Introduction

Both the bifid tongue as well as the medial mandibular and lower lip clefts are extremely rare in any species. [1, 2] These congenital defects are rarely described in horses, as most reported cases are isolated and ignored.[3]

There are several theories on the disease etiology and the involvement of the intrauterine environment, genetic anomalies, and exposure to teratogenic agents are among them.[4] Regarding the genetic aspect, the heritability of orofacial clefts has been more studied in cattle than in horses. Orofacial clefts are reportedly inherited through an autosomal recessive gene. [5] On the other hand, studies in humans have shown that a mutation in the specific locus called interferon regulatory factor 6 is associated with orofacial cleft syndromes. [6]

Furthermore, these developmental anomalies can be lethal, partially lethal, or compatible with life while causing or not only aesthetic defects. [7] In the less severe forms of the disease, only the lower lip is split. However, often, the cleft extends to the mandibular symphysis.[1]

In the literature, the mandibular symphyseal cleft is more described in humans and is characterized by important features such as a complete median cleft of the lower lip with the cleft extension varying from simple to a complete cleft involving the tongue, lower lip, and chin that may extend to the cervical region at variable distances. The mandibular cleft may also have distension that promotes independent movements of the mandible segments, tongue anomaly, including the bifid tongue. There may also be associated characteristics, such as the lack of hyoid bone, thyroid cartilage and even the sternal manubrium.[8,9]

Congenital tongue anomalies occur as an isolated event or may coexist with other anomalies in humans. [10, 11] Among the etiologies highlighted to explain the tongue bifurcation is the persistence of the buccopharyngeal membrane during embryonic development, bands of amniotic constriction in the region of the branchial arches, environmental damage and excess of vitamin A. However, the most likely embryological explanation is a faulty

mesodermal migration for the midline structures of the mandibular portion of the first branchial arch. [12]

The rarity and varying severity of these conditions cause the lack of consensus on the handling and timing of the needed surgical procedures.[13] However, early and simultaneous correction of soft and hard tissues is recommended to facilitate normal growth and function. Unfortunately, there is no long-term follow-up on intervention in these cases to assess mandible growth and evolution. [2]

Case report

A male foal of the Mangalarga Machador breed, 1 month and 15 days old, was treated at the Veterinary Hospital of the University of Brasilia (UnB). The foal presented a congenital malformation of the jaw, lower lip and tongue. The animal born with the malformation resulted from an embryo transfer and, despite apprehending little, was able to suckle on the mare and also showed interest in grass feeding. The foal also coughed but had no runny nose. The mare used as a recipient had neither contact with teratogenic agents nor complications during pregnancy.

Upon physical examination, vital parameters were within the standard for the equine specie while showing the presence of bifid tongue, lower lip and mandible brachygnatism separated in the lower incisors (Figure 1). Subsequently, a radiological examination indicated a faulty union of the median symphysis of the mandible that required surgical intervention (Figure 2 and 3).

Surgical procedure

The foal underwent a surgical procedure with general inhalation anesthesia, in the supine position. Reductive glossoplasty was performed, where the medial aspects of the tongue halves were excised and a new tongue was reconstituted by suturing the parts, layer by layer. A 2-0 polyglactin thread with continuous simple suture was used to join the parts, whereas a nylon 0 thread with simple interrupted suture was used in the external tongue. The lip was excised medially up to the hemimandibles while the tissue was split for better access. In the mandible, the medial ends of the hemimandibles were denuded and curetted. The hemimandibles were joined using two screws (38 mm x 4.5 mm and 36 mm x 4.5 mm) and a cerclage using steel wire (nº 4) was performed (Figure 4). Also, a simple continuous suture using 2-0 polyglactin was performed subcutaneously followed by a simple interrupted suture using nylon 2-0 to close the skin and complete the procedure.

Post operative care

In the postoperative period, the analgesic and anti-inflammatory therapy consisted of meloxicam (0.6 mg/kg, IM, once a day for 10 days), dipyrone (25 mg/kg, IV, twice a day, for 5 days), and ketamine (0.4 mg/kg, daily, SC, four times a day for 5 days). The surgical wound was cleaned with 0.12% chlorhexidine solution three times a day.

During recovery, the animal presented tongue and lower lip edema, which, at first, made swallowing and suckling difficult. Therefore, the mare's milk was administred via tube feeding, a liter of milk every two hours. Dexamethasone (0.1 mg/kg, IV, twice a day, for 3 days) was also administered and a cold compress was applied.

Additionally, because the foal presented leukocytosis (25.5 x 103/ µl) on the second day after surgery, complementary antibiotic therapy with gentamicin (6.6 mg/kg IM,

once a day, for 5 days) was needed. New radiological exams were performed every 3 days, when radiolucent areas were detected close to the screws, indicating rejection.

Eight days after surgery, the animal was sedated to remove the screws and cerclage. A new cerclage was placed fixing the lower incisors (Figure 5). The suture stitches were removed 14 days after surgery. Due to financial limitations, on the eleventh day after surgery, the patient was discharged with the recommendation to clean the mouth with 0.12% chlorhexidine three times a day, while observing the integrity of the cerclage fixed on the incisor teeth (Figure 6). It was also recommended to clean the wounds present in the mandible with a 1% iodine solution.

Finally, 1 tablet of azithromycin (1000 mg) was prescribed once a day for 14 days, 3 tablets of metronidazole (400 mg, every 12 hours for 7 days), dipyrone SID for 5 days, meloxicam 2% SID for 5 days and omeprazole for 14 days.

Outcome

At the time of this report, the animal is still alive, exhibits a good body score, eats well, the quality of life improved after surgery and has a good life expectancy.

Discussion

The causes of most anomalies are unknown. The possible causes are genetic malformation, teratogenic substances, infections, trauma, metabolic and endocrine malformations. [14, 15] In humans, bifid tongue in babies is associated with diabetic mothers, [16] thus highlighting the importance of evaluating the calving mare to avoid repeating the malformation in future pregnancies. Furthermore, it is impossible to make breeding recommendations given the fact that the understanding of genetic inheritance is very

precarious. [3] However, there is a consensus regarding avoiding passing on genetic traits that can harm the well-being and quality of life of future animals.

There is a report of three cases involving mule foals, crossing hybrids between a mare (*Equus caballus*) and a donkey (*Equus asinus*). The animals had mandibular cleft and some level of difficulty to suckle that resulted in weakness either due to the suckling difficulty or the constant loss of milk through the mouth. In the end, two mule foals were submitted to surgical intervention and one died post-surgery. The other two foals were subsequently lost to follow-up. [17]

Another report of a buffalo calf with only a forked tongue describes that the animal also could not suckle milk before the surgical procedure, but responded well to the surgical intervention. [7]

Despite having a cleft mandible and even a cleft tongue, some foals seem to have good suction, enough to survive in the short term. However, these animals may present more problematic long-term survival when solid food is introduced. Therefore, surgical intervention becomes a relevant procedure in such cases. [17]

In humans, simultaneous early correction of hard and soft tissue malformations has become the gold standard for the management of midline cleft. This procedure results in normal anatomical development and mandible growth that contributes to excellent functional capabilities, such as the chewing.[2] It is also possible to maintain normal masticatory function, regardless of the mobility of individual mandibular segments. [18] The foal in the present report maintained its normal chewing functions and was able to feed normally, both liquids and solids.

Compared to the available literature for correcting the mandibular cleft, the reported surgical procedure did not require performing incisor scaling in its medial aspect to allow contact at the mandibular symphysis, as reported for mule foals. [18] However, it is noteworthy that using the stainless-steel wire for cerclage is important and has been highlighted in the literature. [17, 18] Furthermore, using this type of wire requires close monitoring and tightening of the wire loop because of developing instability, which may need to be repeated several times.[17]

The use of cortical screws associated with cerclage, on the other hand, showed better results in both mandibular clefts and mandibular fractures, stabilizing and realigning the mandible. [17, 19] Current study mentions the displacement of pins that could have been avoided with the use of small coaptators at the ends of the pins. [19]

Conclusions

Despite being rare anomalies, there are relevant findings for the prognosis and corrective treatment of these types of malformations. Moreover, it needs to be highlighted that the high degree of difficulty to suckle can represent a reserved prognosis for the animal. As for the treatment, surgical intervention is required so the animal can feed in the long term.

Therefore, by combining the prognosis, satisfactory correction techniques, supportive treatment for the foal and postoperative follow-up, success in treatment can be improved.

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236 Figure 1. Field examination showing bifid tongue and hemimandibles separation.

237 Figure 2. Pre operatory DV radiographic image.

238 Figure 3. Post surgery DV radiographic image showing screws and cerclage placement.

239 Figure 4. DV radiographic image after placement of a cerclage in the ventral incisors.

240 Figure 5. 7th post surgery day. Four days before discharge