Research, Society and Development, v. 9, n. 9, e577997615, 2020
(CC BY 4.0) | ISSN 2525-3409 | DOI: http://dx.doi.org/10.33448/rsd-v9i9.7615

Diprosopia incompleta com inversão ventral do cerebelo em um bovino

Incomplete diprosopia with ventral cerebellum inversion in one bovine calf

Diprosopía incompleta con inversión ventral del cerebel en un ganado

Received: 14/08/2020 | Reviewed: 24/08/2020 | Accept: 27/08/2020 | Published: 30/08/2020

Kaisa Freitas de Araújo
ORCID: https://orcid.org/0000-0001-9834-3594
Universidade Federal de Rondônia, Brasil
E-mail: araujokaisa@gmail.com
Vagno da Silva Miler
ORCID: https://orcid.org/0000-0001-5763-1901
Universidade Federal de Rondônia, Brasil
E-mail: vsmmedvet@gmail.com
Kétury Silva dos Passos
ORCID: https://orcid.org/0000-0002-8205-949X
Universidade Federal de Rondônia, Brasil
E-mail: keturypassos@hotmail.com
Junior Henrique da Silva Moreira
ORCID: https://orcid.org/0000-0002-9426-9312
Universidade Federal de Rondônia, Brasil
E-mail: jr_hsm@hotmail.com
Taisa Fernanda Conceição dos Santos
ORCID: https://orcid.org/0000-0002-9484-0574
Universidade Federal de Rondônia, Brasil
E-mail: taisa_fernanda1@hotmail.com
Sandro de Vargas Schons
ORCID: http://orcid.org/0000-0001-9811-5356
Universidade Federal de Rondônia, Brasil
E-mail: sandroschons@unir.br

Resumo

Este trabalho teve por objetivo relatar um caso de malformação em um bovino recém-nascido, macho, da raça girolando, encaminhado ao Centro de Diagnóstico Animal (CDA) da

Universidade Federal de Rondônia (UNIR) com duplicação craniofacial. Em seguida, realizou-se um estudo descritivo das principais alterações anatômicas observadas durante a dessecação dos músculos da face e remoção do encéfalo. As peças anatômicas foram expostas numa prancha para foto documentação e descrição das malformações. O diagnóstico da diprosopia incompleta foi realizado pela observação das lesões congênitas da face e encéfalo, caracterizados pela junção craniofacial sem separação total da cabeça, apresentando trioftalmia, duplicação das estruturas nasal e bucal com desvio lateral da mandíbula do lado direito. O cerebelo encontrava-se em posição oposta ao normal e se unia aos dois cérebros através dos pedúnculos cerebrais no sentido caudal cranial. Não foi possível determinar a etiologia da malformação, no entanto, ressaltamos a importância da diprosopia na espécie bovina e a necessidade de investigações mais detalhadas sobre os possíveis mecanismos que levam a alterações cranianas.

Palavras-chave: Anomalia congênita; Diprosopo; Duplicação craniofacial.

Abstract

The objective of this case report is to relate a malformation in a newborn male girolando breed bovine calf. The bovine presented craniofacial duplication and was sent to the Animal Diagnosis Center (CDA) at the Federal University of Rondônia (UNIR). The descriptive study of the main anatomical changes observed during the desiccation of the facial muscles and removal of the brain was performed. The anatomical parts were exposed on a board for photo documentation and description of the malformations. The diagnosis of incomplete diprosopia was made by observing the congenital lesions on the face and brain characterized through the craniofacial junction without total head separation; the animal presented triophthalmia, duplication of nasal and oral structures with lateral deviance of the right jaw side. The animal's cerebellum was found in the opposite position and was united with the two brains through the cerebral peduncles in a caudal cranial orientation. In this case report, it was not possible to determine the malformation etiology, however, it emphasizes the importance of diprosopia in bovine and it suggests the need of further studies about the mechanism that produce cranial alterations.

Keywords: Congenital anomaly; Diprosope; Duplicación craneofacial.

Resumen

Este estudio tuvo como objetivo reportar un caso de malformación en un recién nacido, macho, gyrolando bovino que fue enviado al Centro de Diagnóstico Animal de la Universidad

Federal de Rondônia con duplicación craneofacial. Luego, se realizó un estudio escriptivo de los principales cambios anatómicos observados durante la desecación de los músculos cara y la extirpación del cerebro. Las piezas anatómicas se exhibieron en un tablero para documentación fotográfica y descripción de las malformaciones. El diagnóstico de diprosopía incompleta e hizo observando la unión craneofacial sin separación total de la cabeza, con triophthalmos, duplicación de las estructuras nasal y oral con desviación lateral de la mandíbula en el lado derecho. El cerebelo estaba en una posición opuesta a la normal y unía los dos cerebros a través de los pedúnculos cerebrales en dirección caudal craneal. No fue posible determinar la etiología de la malformación, sin embargo, enfatizamos la importancia de la diprosopia en la especie bovina y la necesidad de investigaciones más detalhadas sobre los posibles mecanismos que conducen a cambios craneales.

Palabras clave: Anomalía congénita; Diprosope; Duplicación craneofacial.

1. Introduction

Congenital malformations are structural and/or functional anomalies present in animal birth with an ability to affect a single body structure or function, part of the body system, or the entire system (Node & De Lahunta, 1985). The causes for congenital malformations, in general, are not established; usually they are related to environmental and genetic factors, or the interaction of both (Smith, 2006).

Among malformations, craniofacial duplication or diprosopia occurs when the cephalic region and the facial structures present duplication; in most cases diprosopia presents a monomorphic pattern (Carles et al., 1995). This anomaly can occur in a complete or incomplete form. In the complete form, the duplicated structures have total separation, where the animal is born with a double head, neck, brain, and marrow. In the incomplete form, the structures keep fused (Horovitz, Llerena & Mattos, 2005).

The causes for diprosopia can be attributed to different factors, such as nutrition, teratogens (e.g. radiation), traumatism, the use of corticosteroid, the use of drugs (e.g. benzimidazoles), or the ingestion of toxic plants (Pugh, 2004).

Diprosopia is more common in calves and lambs than cats, dogs, and foals (Özyildiz et al., 2009). The objective of this case report is to describe a case of incomplete diprosopia in a girolando breed bovine calf, as well as to describe morphologic alterations found in the encephalon.

2. Methodology

A newborn male girolando bovine calf presenting a head with face duplication and two fused skulls was sent to the Animal Diagnosis Center (CDA) at the Federal University of Rondônia (UNIR). For this purpose, a descriptive study of the main anatomical changes observed during the physical examination and necropsy was performed. The anatomical parts were exposed on a board for photo documentation and description of the malformations.

The clinical and epidemiological data of the case were filed by the researcher with the farmer, an owner of a dairy farm where 152 male and female cross-bred zebuine bovines were being raised. The cow that delivered the calf with diprosopia was purchased by the farmer. The farmer originally acquired the cow with an existing newborn calf from its first pregnancy. In its first pregnancy on the dairy farm property, which was the second pregnancy of the animal, the cow delivered the calf with diprosopia presented in this case report.

3. Results

During the pregnancy, no abnormal animal behavior was observed, therefore no treatment or different animal management was implemented; however, some animals on the property presented wounds in the udder region that were characteristic of pox. The calf from this case was born from normal parturition.

In the physical exam nasal discharge in the was observed containing mucus and pus, triophthalmia, with two lateral eyes and one located in the middle, between the fused skulls in the zygomatic bone level. Symmetric movements of the lateral eyeballs in each of the faces were also observed; however, the eye in the central portion of the face did not present stimulus to light and had serious fluid discharge. In the central cranial region, there was duplication of buccal and nasal structures with deviance of the lateral right jaw (Figure1).

During the procedure to remove the encephalon, it was observed two skulls fused in the lachrymal bone, zygomatic, frontal, parietal, and temporal region, forming one braincase.

In the dorsal view of the encephalon (Figure 2A), it was observed two telencephalons with four separated cerebral hemispheres. They were separated due to increase of the longitudinal cerebral fissure (LCF), which was more pronounced in the right hemisphere. The caudal portion of longitudinal cerebral fissure became more convex facing the ventral portion of the pons that was in the dorsal portion of the encephalon. In the cerebral hemispheres, it

was observed the metancephalon inversion with the cerebral peduncles and the cerebellum in the ventral portion, and the trapezoid body located dorsally in direction of the cerebellum.

In the ventral view (Figure 2B), it was observed a pronounced separation of the two hemispheres with an abnormal cavity formation between the metencephalon and telencephalon.

Figure 1. Bovine fetus, craniofacial duplication with triophthalmic diprosopus, without complete head separation and lateral deviation of the mandible on the right side, cranial view.

.

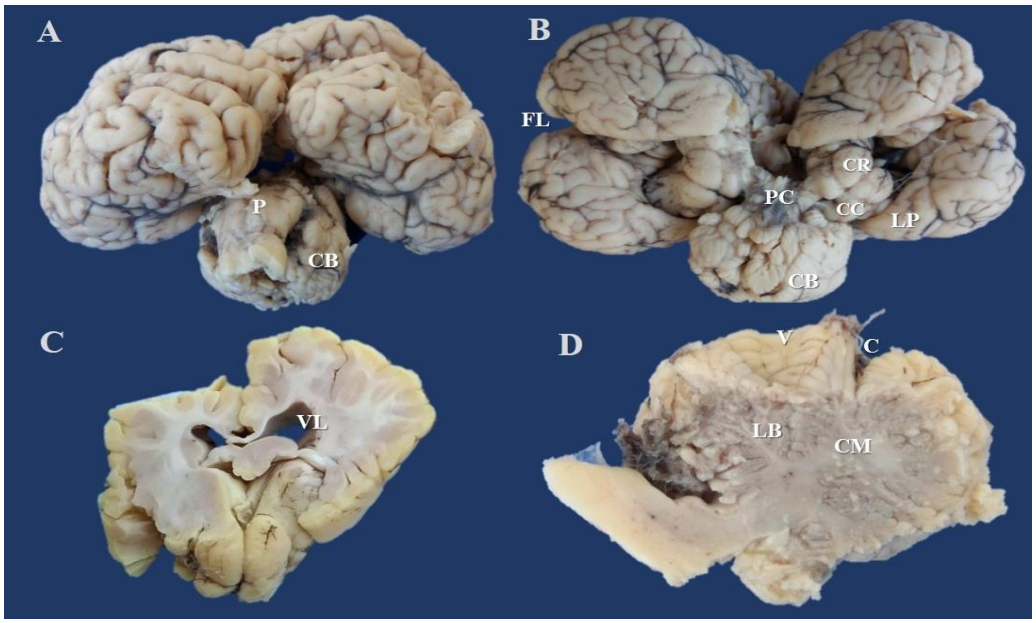
Source: Authors

The pronounced separation was due to absence of pyriform lobes (LP), hypophysis, mesencephalic duct, and optic chiasma separation; it was more pronounced in the right cerebral hemisphere, which led to greater colliculus exposure. In the middle space of the hemispheres, it was possible to visualize the superior colliculus (CR) and inferior colliculus (CC) connected to each hemisphere and carotid artery. The right-side transversal cut of the telencephalon (Figure 2C) showed patterns of normality, while the left side showed a discreet hydrocephalus in the lateral ventriculus (VL).

The cerebellum (Figure 2B) was connected to the telencephalons through a single cerebellar peduncle (PC), with a single pons (P) and four ventricles. In the longitudinal cut of one cerebellum split (Figure 2D), it was observed tapering of white substance (LB), enlargement of cortex (C), underdeveloped medullary body (CM), and cerebellar vermis agenesis.

The left jaw was disjointed in the angle of the transversus menti with both tongues fused in the posterior side; the jaw was nerveless by a single pair of hipoglosso nerves. The lateral parts of the jaw were completely developed; however, they presented an irregular number of premolars and molars. The middle part was rudimental, presenting a cluster of unorganized tissues with premolars and molars.

Figure 2. Bovine encephalon. A) Encephalon formed by two telencephalic hemisphere united with ventral inversion, caudal view. B) Encephalon, central view. C) Encephalon transversal cut, lateral ventricle presenting hydrocephalus. D) Cerebellum longitudinal cut.



Source: Personal collection.

4. Discussion

The diagnostic of incomplete diprosopia was based on macroscopics findings of fusion of two skulls with similar anatomic structures, and duplicated buccal and nasal structures without the duplication of vertebral column, similar to what was described in bovines (Salami et al., 2011; Silva et al., 2010).

The tongue presented duplication in its anterior section and was united in the caudal portion where both manifested swallowing reflex. Those results were also observed by Rotta, Torres & Mota (2008) and Atasever & Ekebaş (2016). The cleft palate is the most common

congenital malformation associated with diprosopia (Saperstein, 1981); however, it was not found in the calf from this case. Nevertheless, it was observed anatomic alterations in the buccal cavity, which were characterized by the lateral deviance of the jaw, similar to what was described by Fisher et al. (1986).

In the post-mortem examination of the cranium, it was observed the complete duplication of telencephalons, and both were caudally connected to the brainstem through the thalamus, and the cerebellum was found in ventral position. Similar alterations were described in bovines; however, the cerebellum was absent (Santos et al., 2005). A triophthalmia is an alteration that is not commonly reported in diprosopia cases. According to Barr (1982), the duplication of eyes and noses implies the duplication of the principal portion of the prosencephalon, which could promote three or four optic vesicles to emerge. The animal in this case did not present cardiovascular malformation. The other systems showed no morphofunctional changes.

In veterinary medicine, there is no pre-disposition for diprosopia by sex or breed; however, in the literature there is a higher frequency of diprosopia in Hereford (Nak et al., 2011; Saperstein, 1981), and Holstein Friesian breeds, according to a study done by Biasibetti et al. (2010), where seven of nine animals were of Holstein breed.

It is not clear what causes diprosopia. However, Smith (2006) explains that this pathology is related to the transference of one mutation gene during the breeding of different breeds. For Dennis and Leipold (1979), diprosopia is caused by a group of malformation in siamese twins, of which could be the result of the fusion of two organisms or portions of bodies partially duplicated. However, Lima (2015), considers that the malformation is excess Sonic Hedgehog (SHH) protein production, responsible for facial characteristics. This protein is essential for the fetus' facial formation, and holds an important role in the growth, cellular specialization, and normal development of the body; the protein is important for neurogenesis and normal nervous tissues, eyes, and body parts. Nevertheless, the causes for the protein increase are not established.

In this case report it was not possible to determine the malformation etiology. This case report showed the importance of diprosopia in bovine and it suggests the needs of further studies about the mechanism that produce cranial alterations.

5. Conclusion

The presence of craniofacial duplication without total heads separation with two skulls fused in the frontal, parietal, and temporal region was the most important factor to the diprosopia diagnostic of the bovine calf in this case.

References

Atasever, A., & Ekebaş, G. (2016). Bir montofon buzağıda diprosopus olgusu. *Ankara Universitesi*. Veteriner Fakultesi Dergisi. 63, 415-419. DOI: 10.1501/Vetfak_0000002763

Barr, M. (1982). Facial duplication. Case review and embryogenesis. *Teratology*, 25,153-159. DOI: 10.1002/tera.1420250205

Biasibetti, E., D'Angelo, A., Bellino, C., Gay, L., Gianella, P., & Capucchio, M. T. (2011). Diprosopia/dicephalia in calves in northern italy: clinical and aetio-pathological features. *Anatomia, Histologia, Embryolgia*. 40, 433–440. DOI: 10.1111/j.1439-0264.2011.01093.x

Carles, D., Weichhold, W., Alberti, E. M., Léger, F., Pigeau, F., & Horovitz, J. (1995). Diprosopia revisited in light of the recognized role of neural crest cells in facial development**.** *Journal of Craniofacial Genetics and Developmental Biology*, 15, 90-97. Retrieved from https://www.researchgate.net/publication/15563084_Diprosopia_revisited_in_light_of_the_recognized_role_of_neural_crest_cells_in_facial_development

Dennis, S. M., & Leipold, H. W. (1979). Ovine congenital defects. *Veterinary Bull.* 49, 233-239.

Fisher, K. R. S., Partlow, G. D., & Walker, A. F. (1986). Clinical and anatomical observations of a two-headed lamb. *The Anatomical Record*, 214, 432-440. DOI: 10.1002/ar.1092140415

Horovitz, D. D. G., Llerena, J. R., & Jr Mattos, R. A. (2005). Atenção aos defeitos congênitos no Brasil: panorama atual. *Caderno de Saúde Pública*, Rio de Janeiro, 4, 1055-1064. DOI: 10.1590/S0102-311X2005000400008

Lima, M., Nunes, A., Bandeira, I., Maciel, J., Freitas, M., & Santos, C. Diprosopia. In: *Encontro de Extensão, Docência E Iniciação Científica (EEDIC)*, 11, 2015, Quixadá: Anais, 2015. ISSN: 2446-6042. Retrieved from http://publicacoesacademicas.unicatolicaquixada .edu.br/index.php/eedic/article/view/648/440

Nak, D., Yilmaz, R., Yilmazbaş, G., & Nak, Y. (2011) A rare case of diprosopus, tetraophthalmus and meningoencephalocele in a lamb. *Kafkas Universitesi Veteriner Fakultesi Dergisi*. 17, 333-336. Retrieved from https://www.researchgate.net/publication/ 268445162_A_Rare_Case_of_Diprosopus_Tetraophthalmus_and_Meningoencephalocele_in_ A_Lamb

Özyildiz, Z., Oral, H., Kurt, B., Yıldız, S., & Güngör, Ö. (2009). İki melez buzağıda diprosopus. *Kafkas Universitesi Veteriner Fakultesi Dergisi*, 15, 305-308. Retrieved from https://www.researchgate.net/publication/261246465_Iki_Melez_Buzagida_Diprosopus

Pugh, D. G.. (2004) *Clínica de ovinos e caprinos*. Ed. Roca. São Paulo.

Rotta, I. T., Torres, M. B. A. M., & Motta, R. G. (2008). Diprosopia em bovino. *Arquivo Brasileiro de Medicina Veterinária e Zootecnia,* 60, 489-491. DOI: 10.1590/S0102-09352008000200032

Salami, O. S., Okaiyeto, S. O., Danbirni, S. B, Ibe C., Allam L., & Kudi A. C. (2011). A case of diprosopus monauchenos in a day old calf (White Fulani× Friesian cross) in an integrated dairy farm. *International Journal* of *Livestock Production*, 2, 55-58. Retrieved from https://academicjournals.org/journal/IJLP/cited-by-article/139ADA91626

Santos, M. S., Adami, M., Oliveira, A. C. G., Souza, M. S., Silva, R. D. G., Pinto, M. G. F., Almeida, A. E. F. S., & Faria, M. M. M. D. (2005). Diprosopo em bezerro (relato de caso). *Revista Brasileira de Saúde e Produção Animal*, 6, 24-30. Retrieved from http://www.rbspa.ufba.br/index.php/rbspa/article/viewArticle/629

Saperstein, G. (1981). Diprosopus in a hereford calf. *The Veterinary Record*, 108, 234-235.

Silva, A. A., Papa, J. P., Fonseca, E. F., Ferreira, J. C., Ignácio, F. S., Stelmann, U. J. P., & Pompermayer, L. G. (2010). Diprosopus em bezerro - Relato de caso. *Revista Científica Eletrônica de Medicina Veterinária*, 14, 8. Retrieved from http://faef.revista.inf.br/imagens_arquivos/arquivos_destaque/IE41SIYrKdrAva5_2013-6-25-14-58-10.pdf

Smith, B. P. (2006). *Medicina Interna de Grandes Animais*. (3a ed.), Barueri, São Paulo. Manole, 1465-1469.

Percentage of contribution of each author in the manuscript

Kaísa Freitas de Araújo – 40%

Vagno da Silva Miler – 10%

Kétury Silva dos Passos – 10%

Junior Henrique da Silva Moreira– 10%

Taisa Fernanda Conceição dos Santos– 10%

Sandro de Vargas Schons – 20%