Bovine thoracic Ectopia cordis in southeastern Rio Grande do Sul, Brazil

Ectopia cordis torácica bovina no sudeste do Rio Grande do Sul, Brasil

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ABSTRACT  
This study describes and illustrates a case of thoracic ectopia cordis (EC) in a cross-bred calf in the southeastern region of Rio Grande do Sul, Brazil. A cow suffering from dystocia due to fetal static gave birth to a calf with the heart and pericardial sac outside the thoracic cavity and respiratory distress. The animal survived for 40 min after giving birth. EC is defined as the complete or partial displacement of the heart out of the thoracic cavity. It is a rare birth defect in the fusion of the anterior chest wall, which results in the extrathoracic location of the heart. Three possible theories for the development of this condition have been described in animals: the first is related to a primary failure in the descent of the heart and fusion of the midline of the body, the second is associated with rupture of the amnion, and the third is associated with amniotic band syndrome. Toxic plants, inbreeding, and some pharmacological compounds have been associated with the development of the anomaly. However, in most cases, the precise cause of the appearance of the malformation is unknown. This animal was considered a sporadic case of EC. EC has been described more frequently in cattle, although there are reports in other species, such as dogs, cats, and sheep. Treatment can be surgical, but this procedure is rarely an option.

Keywords: calf, congenital anomaly, heart, ruminants, neonatology.

RESUMO  
Este estudo descreve e ilustra um caso de ectopia cordis torácica (EC) em um bezerro de raça cruzada na região sudeste do Rio Grande do Sul, Brasil. Uma vaca que sofria de distocia devido à estática fetal deu à luz um bezerro com o coração e o saco pericárdico fora da cavidade torácica e aflição respiratória. O animal sobreviveu por 40 minutos após o parto. A CE é definida como o deslocamento total ou parcial do coração para fora da cavidade torácica. É um raro defeito congênito na fusão da parede torácica anterior, o que resulta na localização extratorácica do coração. Três teorias possíveis para o desenvolvimento dessa condição foram descritas em animais: a primeira está relacionada a uma falha primária na descida do coração e fusão da linha média do corpo, a segunda está associada à ruptura do amnion e a terceira está associada à síndrome da banda amniótica. Plantas tóxicas, endogamia e alguns
compostos farmacológicos foram associados ao desenvolvimento da anomalia. No entanto, na maioria dos casos, a causa precisa do aparecimento da malformação é desconhecida. Este animal foi considerado um caso esporádico de EC. A EC tem sido descrita mais frequentemente em bovinos, embora existam relatos em outras espécies, como cães, gatos e ovelhas. O tratamento pode ser cirúrgico, mas este procedimento raramente é uma opção.

**Palavras-chave:** bezerro, anomalia congênita, coração, ruminantes, neonatologia.

### 1 INTRODUCTION

Congenital cardiovascular anomalies are abnormalities present from birth and may arise due to a combination of genetic, environmental, infectious, toxicological, pharmaceutical, nutritional, or other influences or a combination of these factors (DANTAS et al., 2010).

The prevalence of congenital heart anomalies in cattle is not as well defined as in other animal species. Congenital heart defects in this species have been suggested to have a prevalence of 0.2–2.7% (CAIVANO et al., 2023).

Ectopia cordis (EC) is a rare congenital anomaly with a high mortality rate, characterized by a defect in the fusion of the anterior chest wall that results in an extrathoracic location of the heart (JEZEK et al., 2016). This condition can be classified as total or partial according to the volume of the exteriorized heart (JEZEK et al., 2016).

In this context, this study aimed to describe and illustrate a case of thoracic EC in a cross-bred calf in the southeastern region of Rio Grande do Sul, Brazil; thus, contributing to the literature and helping other veterinarians with the diagnosis of this disease in the future. Furthermore, it is proposed to write narrative review of the literature on EC in cattle to promote a better understanding of the pathogenesis of this disease.

### 2 CASE REPORT

A cow that had been in dystocia labor for 2 days, caused by fetal static, was treated in the municipality of Cerrito, the southeastern region of Rio Grande do Sul (RS), Brazil (Figure 1A). The fetus was in a posterior longitudinal position
and a superior position with hip flexion. A maneuver was performed to correct the flexion of the hip joint to allow its traction.

At birth, the animal had the heart and pericardial sac completely externalized in the thoracic region due to a sternal cleft and a tegumentary defect (Figure 1B - inset). The calf had difficulty breathing and died 40 min after birth. The owner did not allow a necropsy to be carried out; therefore, no other anatomical changes could be observed. The herd to which the mother belonged had no control over inbreeding, and the source of her food was the native vegetation of the field.

Figure 1. (A) Rio Grande do Sul state, location in Brazil, showing the seven mesoregions and location of the case. (B) Thoracic ectopia cordis in the calf with an externally visible heart. The inset shows a closer view of the heart with a pericardial sac that covers the organ.

3 DISCUSSION

The association of history, clinical signs, and anatomopathological findings allowed the diagnosis of thoracic EC.
Congenital heart diseases are usually due to damage to cells or genes that are responsible for cardiac development in the fetal period (MORAES et al., 2014). Three possible theories have been mentioned for the development of EC in animals as follows: the first is related to a primary failure in the descent of the heart and the fusion of the midline of the body. The development of the ventral body wall begins approximately the 8th day of embryonic life with differentiation and proliferation of the mesoderm followed by its lateral migration. The heart originally develops in a cephalic location and reaches its final position by lateral bouncing and ventral flexion of the embryo on approximately the 16th to 17th day. The midline fusion and formation of the thoracic and abdominal cavities are completed by the 9th week of the embryonic period. The complete or incomplete failure of midline fusion at this stage results in disorders ranging from isolated EC to complete ventral evisceration. The second theory is associated with amnion rupture, in which, during fetal development, the amnion surrounding the embryonic structures ruptures and the fibrous and sticky bands of this structure become entangled in the developing embryo and cause an interruption in the development of parts of the embryo or fetus that can lead to various deformities such as EC, midline sternal cleft, frontonasal dysgenesis, and limb deformities (SHAD; BUDHWANI; BISWAS, 2012). The third theory is associated with amniotic band syndrome. This syndrome is characterized by birth defects with various clinical manifestations, including EC. Its etiology is unknown and two pathophysiological mechanisms for its development are proposed. In the first, there is an early rupture of the fetus with the amnion that would allow the contact of the amniotic chorionic surface, resulting in the adhesion of this structure to several fetal segments, which would cause the fibrous bands to imprison the fetal body. The second proposed mechanism establishes that a defect occurs in the germ plasma with vascular rupture and alteration of morphogenesis during the gastrulation phase and the appearance of the defect (SHAD; BUDHWANI; BISWAS, 2012).

EC cases are usually associated with other cardiac anomalies, such as ventricular septal defects, coronary changes, and persistent ductus arteriosus.
EC has also been associated with other systemic congenital anomalies, such as abdominal wall defects, cranial and facial malformations, cleft lip and palate, anencephaly, hydrocephalus, neural tube defects, pulmonary hypoplasia, genitourinary malformations, and gastrointestinal defects (ERÖKSÜZ; METIN; ERÖKSÜZ, 1998). It should be noted that in this study, the owner of the animal did not allow the veterinarian to perform the necropsy, making it impossible to visualize other possible defects that the animal could present. It is important to remember that necropsy contributes to the body of scientific knowledge by increasing our understanding of anatomy and physiology in health and disease, complements clinical medicine, alerts us to the presence of diseases that may be transmissible to other animals (or humans), and guides treatment decisions for at-risk individuals, and in some cases, necropsy findings can give comfort or closure to the owner, especially in the case of a seemingly sudden or unexplained death (KING, 1989).

EC has been described more frequently in cattle, although there are reports in other species, such as dogs, cats, and sheep (HIRAGA et al., 1993). Furthermore, it can be classified as cervical, cervicothoracic, thoracic, thoracoabdominal, and abdominal according to the location of the ectopic heart (MORAES et al., 2014). In this case, the animal had thoracic EC. In human medicine, most cases of EC occur in the thoracic and cervicothoracic regions (SHAD; BUDHWANI; BISWAS, 2012). However, in a study on eight bovines with EC, the authors observed that the most affected region in this species was the cervical region, followed by the thoracic and abdominal regions (HIRAGA; ABE, 1986).

The Holstein Friesian is the most affected bovine breed, but it has already been described in other breeds such as Shorthorn, Hereford, Angus, Guernsey, Simmental, and Limousin (JEZEK et al., 2016). In this case, the animal was a cross-bred bovine from a small rural property in the southeast of RS, where the producer does not control the crossings, which can favor the inbreeding of the herd and, consequently, the appearance of defects transmitted by recessive genes. The precise cause of congenital heart anomalies in cattle, as in all other
animal species, is unknown. It is important to emphasize that the use of teratogen
drugs and toxic plants may be involved in the development of EC (BLUE et al.,
2012). However, no possible toxic plants were observed on the property and the
producer claims that the cow had not been treated for any disease during the
gestational period. This malformation is believed to occur sporadically and is not
associated with a specific cause.

The differential diagnosis of EC is a pentalogy of Cantrell, amniotic band
syndrome, and cylosomas. All these anomalies are considered rare in human
medicine and result in the failure to close body cavities (SHAD; BUDHWANI;
BISWAS, 2012).

4 CONCLUSION

Therefore, this study describes a case of EC and draws attention to the
importance of recognizing and reporting the occurrence of congenital anomalies
in animals by veterinarians, who should seek to determine the possible etiologies
involved in these anomalies to develop efficient prevention and control programs
in Brazilian herds.

ADDITIONAL FILE

Thoracic ectopia cordis in the calf. Observe the heartbeat with the heart located
outside the thoracic cavity. [https://youtu.be/bmeVbc9j-58]

ACKNOWLEDGMENTS

This study was financed in part by the Coordenação de Aperfeiçoamento de
Pessoal de Nível Superior – Brasil (CAPES) – Finance Code 001, by Conselho
Nacional de Desenvolvimento Científico e Tecnológico (CNPq), and by
Fundação de Amparo à Pesquisa e Inovação do Espírito Santo (FAPES).
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