CASE REPORT

F.U. Vet. J. Health Sci.
2019; 33 (3): 207 - 209
http://www.fusabil.org

A Case of Pulmonary Hypoplasia and Anasarca Syndrome with Palatoschisis in a Calf Fetus *

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Received : 27.11.2019
Accepted : 10.12.2019

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Pulmonary hypoplasia and anasarca syndrome is a lethal genetic disorder of cattle characterized by excessive subcutaneous edema, undeveloped lungs and fetal hydrops. In this case report, a calf fetus delivered by the cesarean section from a two-year-old Simental breed heifer was presented. Intense fluid was collected between the subcutaneous tissues of the extracted calf. Pathologically, it was found that the animal had diffuse and severe subcutaneous edema, total palatoschisis in the upper palate, the lungs were smaller than normal, atelectatic and hypoplastic, and the heart was bilaterally dilated. Microscopic examination revealed significant histopathologic findings such as epicardial and endocardial edema in the heart and lungs, endocardial fibrosis, fetal atelectasis in the lungs, and perivascular and interstitial edema. This is the first report in Turkey characterized by anasarca and malformation, suggesting this is a rare case in animals.

Key Words: *Anasarca, fetus, palatoschisis, pulmonary hypoplasia*

Bir Buzağı Fetüsünde Karşılaşılan Pulmoner Hipoplazi ve Anasarka Sendromu ile Palatoşizis Olgusu

Pulmoner hipoplazi ve anasarka (PHA) sendromu, subkutan ödem, gelişmemiş akciğerler ve fetal hidropsi ile karakterize sığırların öldürücü bir genetik bozukluğudur. Sunulan olguyu 2 yaşlı Simental ırkı bir düveden sezaryen operasyonu ile alınan buzağı fetüsü oluşturdu. Çıkarılan fetüsün deri altı dokuları arasında yoğun sıvı toplandığı görüldü. Patolojik olarak hayvanda yaygın ve şiddetli deri altı ödem, üst damakta total palatoşizis, akciğerlerin normalden küçük hacimli, atelektatik ve hipoplastik, kalbin ise çift taraflı dilate olduğu tespit edildi. Mikroskobik incelemede kalp ve akciğerde epikardiyal ve endokardiyal dokuda ödem, endokardiyal fibrozis, akciğerlerde ise fetal atelektazi ile perivasküler ve intersitisyel ödem gibi önemli histopatolojik bulgular gözlendi. Sonuç olarak bu olgunun, hayvanlarda nadir olarak gözlenmesi, anasarka ve anomali ile karakterize Türkiye'de bildirilen ilk vaka olması nedeniyle vakanın bildirimi yapılmıştır.

Anahtar Kelimeler*: Anasarka, fetüs, palatoşizis, pulmoner hipoplazi*

Introduction

Fetal hydrops is defined as excessive fluid accumulation characterized by caverns formed in fetal extravascular compartments and body cavities, placental enlargement, pericardial and pleural effusion and ascites (1-3). Non-immune fetal hydrops is a feature of many genetic diseases and is associated with alpha thalassemia (1). This may also be accompanied by specific abnormalities such as hereditary "Bulldog" calves, especially in Dexter cattle (4, 5). Fetal hydrops has been described in humans as well in the veterinary literature; however, although various studies have been conducted, its etiology remains unclear (1).

Pulmonary hypoplasia and anasarca (PHA) syndrome is a lethal genetic disorder of cattle characterized by excessive subcutaneous edema, underdeveloped lungs and fetal hydrops (5-7). Rare fetal hydrops and PHA have been reported to be recessively inherited in Australian Dexter, Belted Galloway, Maine-Anjou and Shorthorn cattle (5, 6, 8). In addition to cattle affected by PHA, this has also been found in a sheep with similar lesions (9). Furthermore, it has been reported as an autosomal recessive fetal hydrops case in Spanish sheep (10).

In this article, a case of PHA syndrome in a calf fetus delivered by cesarean section from a Simmental heifer was defined and the clinical, macroscopic and pathological findings are presented.

Case Report

In this rare case, we present a calf delivered by cesarean section from a two-year-old Simmental heifer, which was referred to the Fırat University Faculty of Veterinary Medicine Hospital Obstetrics and Gynecology Clinic with long-term pain and dystocia difficulties. In the clinical examination, a calf with anomalies was diagnosed and a cesarean section was planned. During the caesarean section, the right horn of the uterus was observed to be enlarged, the amount of fluid was plenty and intense fluid accumulation between the subcutaneous tissues was observed in the delivered fetus. In

* Turkish Veterinary Gynecology Association VIII. National & II. International Congress, 10-13 October 2019, Antalya/TURKEY

the necropsy, it was first noted that the calf was too large and swollen (Figure 1). Diffuse and severe subcutaneous edema was observed. Caverns of different diameters and sizes contained edema in subcutaneous tissues (Figure 2).

A 40×30×30 cm cystic formation was found on the back of the neck, which was independent of the brain, starting from the first cervical vertebra, located subcutaneously, with clear yellow liquid content (Figure 3). Complete palatoschisis was observed in the upper palate (Figure 2). The abdominal and thoracic cavities were filled with approximately 3 liters of serosanguinous fluid. When the thoracic cavity was opened, the lungs were found to be smaller than normal, atelectatic and hypoplastic and weighing 35 grams (Figure 4). The examination of the heart revealed that it weighed 145 grams and had a globular appearance, showing bilateral dilatation. There were defects in both the atrium and the ventricular septum. Partial atrial defect and ventricular septal defect anomalies were also observed (Figure 5). No significant findings were found in the brain or other organs.

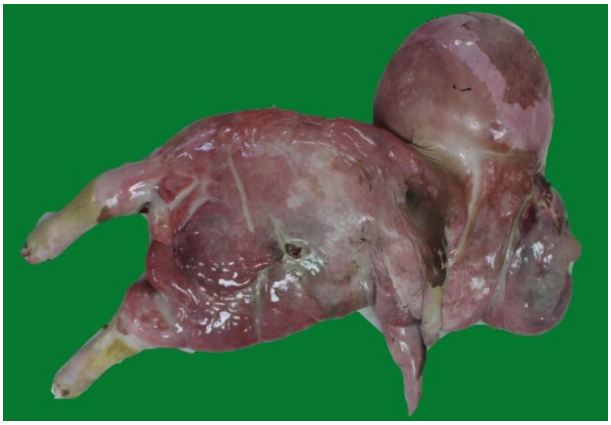

Figure 1. General view of the calf

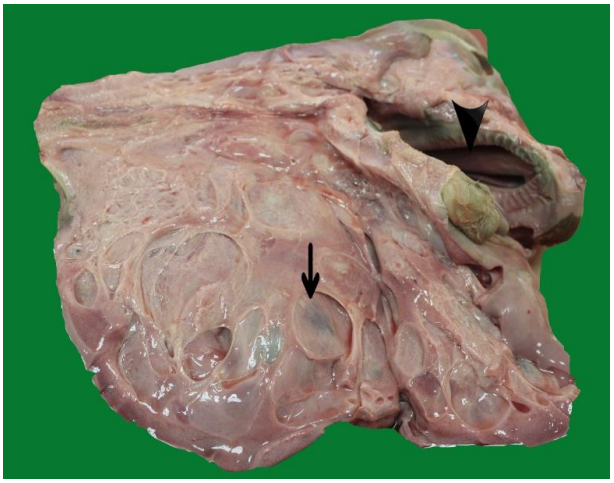

Figure 2. Subcutaneous caverns arrow and palatoschisis arrowhead caused by diffuse and severe edema

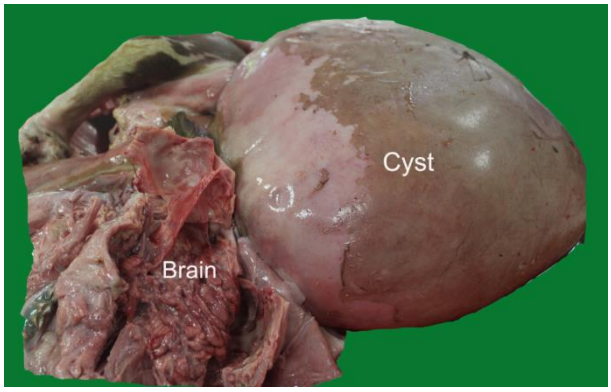

Figure 3. Cyst and autolytic brain seen in the nape of the calf

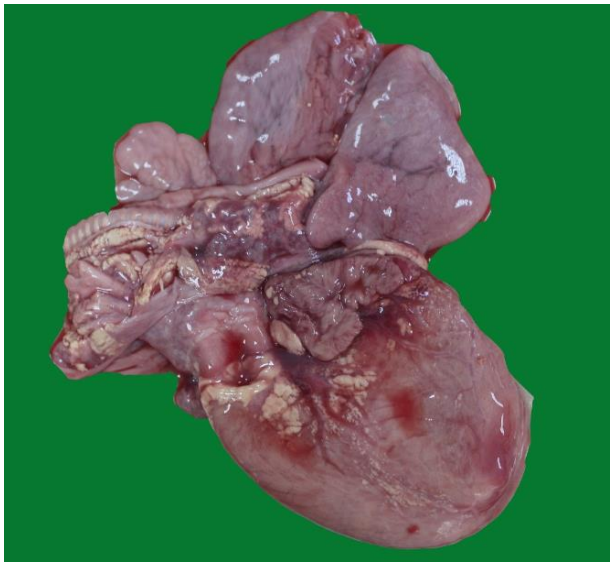

Figure 4. Hypoplastic and atelectatic lungs

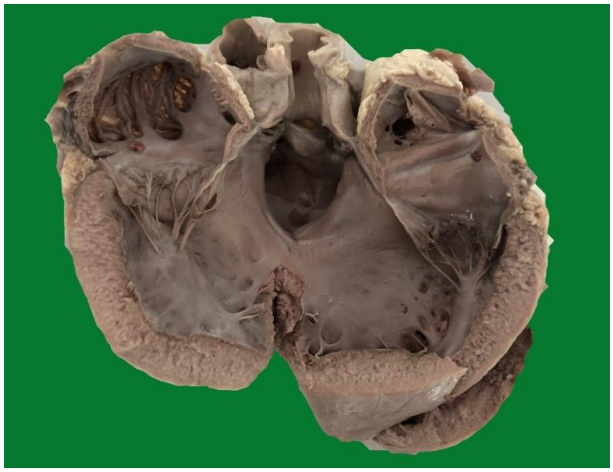

Figure 5. Opened view of the ventricles of the heart undeveloped interventricular septum

Microscopic examination revealed important findings in the heart and lungs. The heart muscle cells were separated from each other due to edema; the transverse lines were prominent and hypertrophic in one place or another.

Randomly distributed focal hemopoietic foci were found in the myocardium. Edema was observed in the epicardial and endocardial tissue. Fibrosis areas consisting of connective tissue and fibers were found in the endocardium (endocardial fibrosis). Moderate edema were noted as well as the hydropic degeneration of papillary muscle. In the lungs, there was no findings except for fatal atelectasis and perivascular and interstitial edema.

Discussion

In the literature review, we came across some similar case reports from other countries, but there is no reported PHA case from Turkey. In a study by Windsor et al. (5) in one of the three different cases they encountered, subcutaneous edema and severe diffuse anasarca type edema were noticed in the calf delivered by cesarean section and protrusion of the tongue was reported. Histopathological examination of the organs revealed that the heart, the thymus and the connective and adipose tissue were identified; however, pulmonary tissue, bronchial or tracheal tissue elements were not identified. In the second case, spongious edema was determined in the lower abdomen and no lung tissue was found in the thoracic cavity. In the third case, moderate anasarca-style edema due to accumulation of subcutaneous fluid was reported. Unlike the other two calves, there was rudimentary lung tissue in the thoracic cavity in this case. Histopathological examination of the tissues revealed significant thickening in the pleural surface, interalveolar and peribronchial areas and the bronchiolar submucosa of the lungs due to edema. There were diffuse hydropic changes in the renal tubular epithelium of the kidney. Alleaume et al. (9) reported a lamb with increased body weight, generalized subcutaneous fluid accumulation and significant effusions in the thoracic and abdominal cavities. The lamb diagnosed as 'fetal hydrops' was reported to have smaller lungs than normal. One of the lambs had a placenta that was severely edematous. Other than these findings, they did not report any other changes or lesions in the remaining organs. Monteagudo et al. (10) diagnosed ‘fetal anasarca’ in lambs that demonstrated an overall increase in body weight and size. They reported that the fetus with edematous tongue had a fragile structure and there was an accumulation of yellow liquid in the body cavities. The authors stated that peripheral and internal lymph nodes were deficient in these cases. On microscopic examination, they detected edema in the soft skin tissues and there was no sample belonging to lymphoid tissue and the lymph nodes were missing; furthermore, they showed extramedullary hematopoiesis in the liver at various severities. In the case reported by Agerholm and Arnbjerg (8) they reported signs of pulmonary hypoplasia, interventricular septal defect, palatoschisis and bilateral cryptorchidism in a Belted Galloway breed calf. Svara et al. (3) reported multiple edematous cysts in the neck, hypoplastic lung and cardiac dilatation in two Cika breed calves diagnosed with PHA, but found no evidence of anomalies. In our case, the macroscopic and microscopic findings showed palatoschisis, atrial defect, ventricular septal defect and lung pathology as well as fetal hydrops; however, there was no cryptorchidism or significant abnormality in the other organs.

In conclusion, this is the first case reported from Turkey characterized by rarely observed anasarca type edema and other abnormalities among animals. We believe that this case will contribute to further researches on this issue and comprehensive investigations are needed to fully understand the etiology of PHA.

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