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CASE REPORT

Ectopia Cordis Cervicalis and Its Surgical Treatment in A Holstein Calf [1]

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Summary

Ectopia cordis is a rare congenital anomaly characterized by partial or complete displacement of the heart, outside the thoracic cavity. Ectopia cordis is usually associated with other multiple anomalies and intra-cardiac defects due to developmental problems in the ventral body wall. In this report, a 6 day-old, 55 kg male Holstein calf with a case of ectopia cordis cervicalis and its surgical treatment has been presented. Following examinations (clinical, haematological, ECG, x-ray, USG and CT) a single staged surgery was performed to correct localisation of the heart in the thoracic cavity. The case was followed up 10 days postoperatively. However, the calf died at the end of post-operative day 10 due to septic shock.

Keywords: *Calf, Ectopia cordis cervicalis, Congenital abnormality*

Holstein Irkı Bir Buzağıda Ectopia Cordis Cervicalis Olgusu ve Operatif Sağaltımı

Özet

Ectopia Cordis, kalbin göğüs kafesi dışında, parsiyal ya da tam olarak anormal yerleşimiyle karakterize, seyrek rastlanan kongenital bir anomalidir. Ectopia cordis, ventral karın duvarının gelişimindeki problemlere bağlı olarak genellikle intrakardiyak defektlerle ya da diğer çoklu anomalilerle ilişkili olabilir. Bu makalede, 6 günlük, erkek, 55 kg ağırlığında, Holstein bir buzağıda görülen ectopia cordis cervicalis olgusu ve operatif sağaltımı anlatılmaktadır. Yapılan muayeneler (klinik, hematolojik, EKG, radyografik, ultrasonografik, BT) sonucunda hastaya, kalbin göğüs kafesi içine alınmasına yönelik olarak tek seansta, cerrahi sağaltımda bulunuldu. Hastanın, 10 gün süreyle postoperatif takibi yapıldı fakat buzağı 10. günün sonunda septik şok nedeniyle öldü.

Anahtar sözcükler: *Buzağı, Ectopia cordis cervicalis, Konjenital anomali*

INTRODUCTION

Ectopia cordis (EC) is the partial or complete displacement of the heart in an abnormal position outside the thoracic cavity [1-3]. While partial or complete ectopia cordis may occur depending on the volume of heart outside the thoracic cavity [4,5], three further different types may be present depending on localisation: cervical (upper and lower cervical ectopia cordis), pectoral and abdominal ectopia cordis [6]. Most frequently seen is the cervical type (82%). Rates at which pectoral and abdominal types are seen are 14% and 3%, respectively [2,3,6]. Observed in 1 in 5.2-7.2 million humans [7]; in animals, this anomaly is most frequently seen in calves [8]. Occurring less frequently than in humans, this anomaly has also been observed in animal species such as horses, goats and pigs [9-11]. Life expectancy in animals with ectopia cordis may range from 3 minutes to several years [5]. Cases where the displacement is either

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through the sternum or the costae usually end with neonatal death. In other types of displacement, animals may live for longer periods [8].

In this report, a case of ectopia cordis in a six-day old calf, in addition, the operation technique and necropsy findings of this condition are described.

CASE HISTORY

This case consisted of a 6-day old Holstein calf weighing 55 kg, with ectopia cordis cervicalis, referred to the Istanbul University Surgery Department. Anamnesis revealed that the calf had been born naturally and on inspection it was observed that it had completed its growth, the skin was complete and the coat was normal. However, during physical examination of the patient, right torticollis and a distinct swelling on the caudal aspect of the left cervical region was identified. The calf had a body temperature of 38.2°C and the lymph nodes and mucosae appeared normal. As well as the evident tachypnoae and tachycardia, moist rales were also audible on lung auscultation. Haemogram findings were assessed to reveal thrombocytopenia and leucopenia. Blood biochemistry revealed that ALP and iron binding values were increased, while LDH, cholesterol, total protein and Ca values were decreased. When electrocardiac findings were examined, heart frequency was found to be 250/min. Also, despite the absence of the P wave in all derivations, the R-R interval was seen to be constant. Supraventricular tachycardia was identified in the patient. Radiodiagnostic examination showed that, as a result of serial radiography and spiral BT assessment directed at the thorax, the base of the heart was at the level of the tracheal bifurcation, higher than normal and that it was located at the level of the jugulum (*Fig 1-A, 1-B* and *1-C*).

Following determination of the patient's clinical, radiographic and electrocardiographic findings, surgical intervention was decided in order to relocate the heart, which was positioned in the neck, to within the thoracic cavity. After the usual pre-operation preparations, the patient was placed in dorsal recumbency and general anaesthesia was induced via mask induction using Isoflurane at a concentration of 4%. The maintenance dose for anaesthesia was 2% Isoflurane (Forane, Abbout®, UK). Balanced fluid electrolyte solution (Lactated Ringer 10 ml/kg/h) was administered to the patient intravenously throughout the operation. Prophilactically, Cephazolin sodium (Sefazol, Mustafa Nevzat® Turkey) was given at a dose of 25 mg/kg IV. The incision was made for a median sternotomy, continued across the region where the heart was positioned and extended towards the ventral aspect of the neck (*Fig 2-A* and *2-B*). Thoracal retractors were placed in the site. It was not possible to separate the heart, which was located immediately under the skin, from adhesions and muscles, therefore pericardiectomy was performed *(Fig 3)*. In order to allow the heart to find its normal anatomic

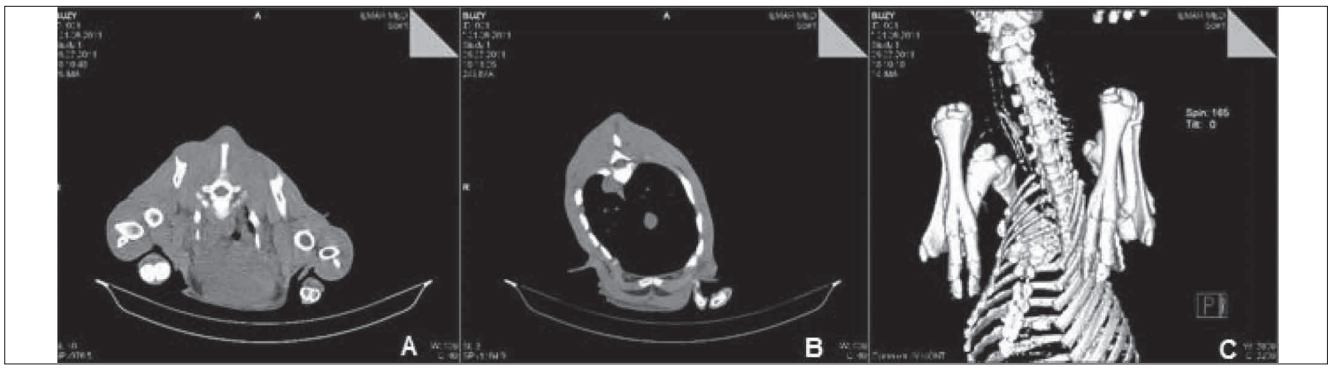

Fig 1. The base of the heart was at the level of the tracheal bifurcation **(A)**, the heart is not seen in thorax cavity **(B, C)**
Şekil 1. Kalp tabanı tracheal bifurkasyonu düzeyindedir **(A)**, göğüs boşluğunda kalp izlenmemektedir **(B, C)**

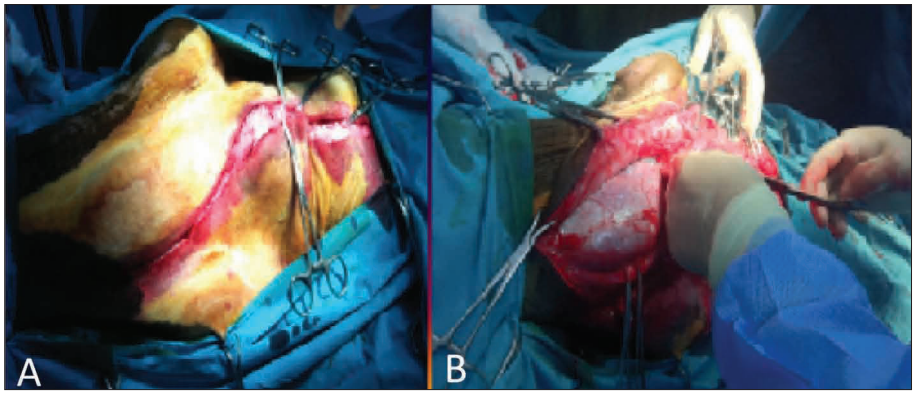

Fig 2. In dorsal recumbency, the incision was made on the ventral aspect of the neck and median sternotomy was performed **(A)**, the heart was located under the skin within adhesions and muscles **(B)**

Şekil 2. Sırtüstü yatış pozisyonunda boynun verntralinden yapılan ensizyonu takiben median sternotomi uygulanmış **(A)** ve kalp açığa çıkarılmıştır **(B)**

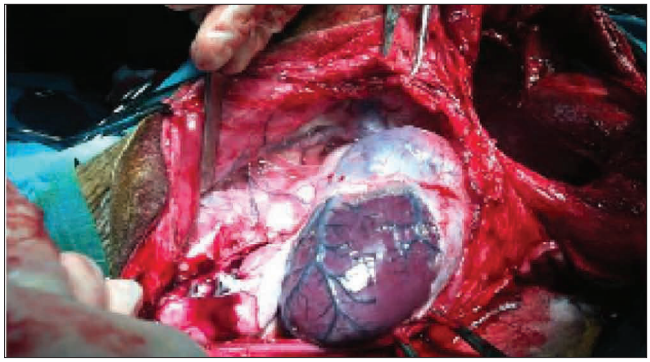

Fig 3. Pericardiectomy site
Şekil 3. Perikardiyektomi bölgesi

location, lobectomy was performed on the cranial lobe of the right lung *(Fig 4-A)*. The heart was then placed in the direction of the lobectomy site *(Fig 4-B)*. The pericardium was not sutured. Sternebrae were brought closer together using cerclage wire and the operation site was closed surgically. During this time, care was taken to intensively ventilate the lungs.

The patient regained consciousness shortly after the operation and was hospitalized. Approximately 24 h after the operation the patient was given food orally. There was no abnormality in the postoperative *(Fig. 7)* clinical findings up until the end of day 8. However, in the following days it was seen that the patient's appetite had declined, body

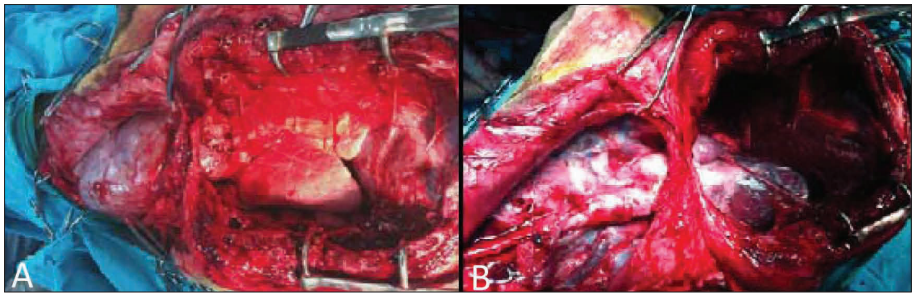

Fig 4. The position of the heart before the right cranial lobectomy **(A)**, the heart's new location after the lobectomy **(B)**

Şekil 4. Kraniyal lobektomi öncesinde kalbin pozisyonu **(A)**, lobektomi sonrasında kalbin yeni konumu **(B)**

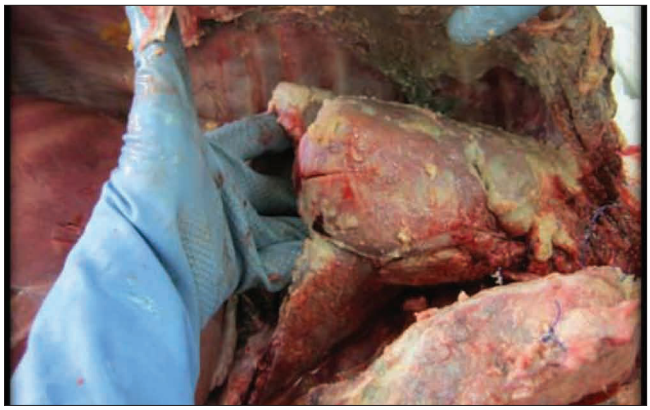

Fig 5. Fibrinous and purulent pleuritis in necropsy findings
Şekil 5. Nekropside fibrinöz ve purulent plöritis bulguları

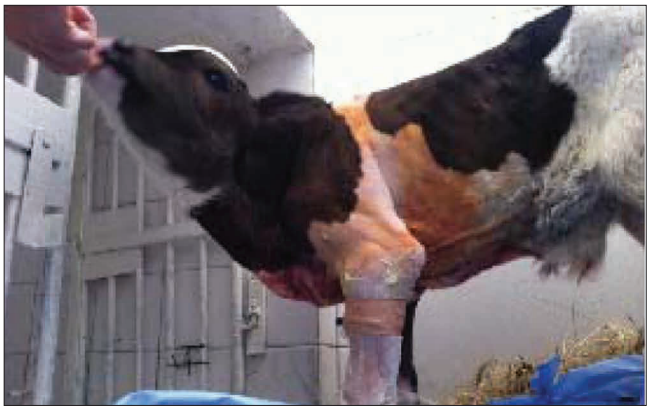

Fig 7. The calf's postoperative period
Şekil 7. Buzağının postoperatif dönemi

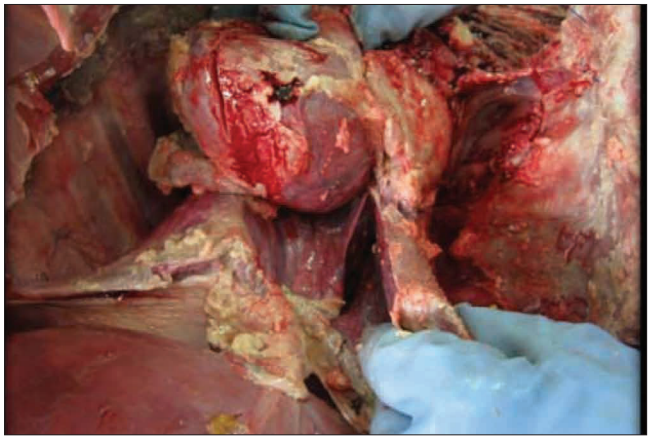

Fig 6. Branching type similarity to pig cattle model
Şekil 6. Domuz-köpek modeline benzer dallanma şekli

temperature had risen to 41°C and leucocytosis was present in haemogram findings. Meanwhile, a sticky seromucous discharge was seen to be present at the incision site. The antibiotic drug Cefazolin HCL given at a dose of 25 mg/kg postoperatively was changed to Ceftiofur hydrochloride (Excenel, Pfizer®) at a dose of 2 mg/kg. However, despite all interventions, the patient died of septicaemia at the end of day 10.

As a result of necropsy findings, the reason of death was found to be fibrinous and purulent pleuritis *(Fig. 6)* and septicaemia.

DISCUSSION

Abnormalities seen in a body structure or function at

birth are called congenital abnormalities [12]. Congenital abnormalities are multidisciplinary cases. These abnormalities may affect a single organ system or structure or all systems or even some systems (cardiovascular or integumentary system) [3,13].

Many factors, such as chemical teratogens, genetic or chromosomal abnormalities, environmental radiation and infectious agents, play a role in the development of congenital defects in calves [3,14]. The conceptus may encounter harmful factors at different developmental stages such as the preimplantation, embryonic or foetal stages [3].

Delayed descension of the heart in the embryonic period is thought to be the mechanism responsible for cervical ectopia cordis [4,5]. It is thought that the factor causing ectopia cordis affects the foetus before day 36 of the pregnancy [4]. While the etiology of this anomaly is not completely understood, it has been reported that this condition is encountered more often in humans and three theories have been suggested for its pathogenesis. These theories are: 1) A primary fault in the process of the lateral body curves meeting at the midline and descending, 2) A fault in midline closure in relation to premature separation of the chorion or the egg, 3) Amniotic band syndrome [5].

Any type of ectopia cordis may be complicated with other malformations either cardiac or non-cardiac [2]. In humans, ectopia cordis may be related to the quintet syndrome; abdominal wall, sternum, diaphragm, pericardium and heart defect, mostly described as Cantrell's pentalogy [7,15,16]. Unusual defects together with ectopia cordis may be also observed in calves. The presence of ectopic lungs and extremities together with ectopia cordis has been reported in a calf with multiple congenital anomalies [17]. In another calf monitored for a long time, ectopia cordis together with tricuspid valve dysplasia was identified and it was thought that this could be related to the Ebstein anomaly however it has been reported that this calf with ectopia cordis cervicalis completed its normal development, became pregnant and calved naturally [18].

Other anatomical defects frequently encountered are; double apex, double vena cava, double vena azygous and sternum abnormalities. In veterinary sources, while branching of the heart along the arcus aorta has been recorded, branching in pigs, dogs and cattle have been reported in three models [4].

Although branching of the blood vessels in calves with ectopia cordis cervicalis is in the type of branching mostly seen in dogs or similar to hybrid branching seen in pig and dog combination, the branching type in the necropsy findings of our case shows similarity to the pig-cattle model *(Fig. 5)*. Among characteristic findings in calves with cervical ectopia cordis are listed; a shortened and widened manubrium, xyphoid cartilage hypoplasia, double appearance sternebrae and presence of multiple sternebrae [4].

In calves with ectopia cordis cervicalis, as a result of surgical treatment to move the heart into the chest cavity, no literature has been found stating that the patient had survived for more than several hours.

The patient in this case report tolerated the operation well. The reason for septic shock development in this case was thought to be due to the pre-existing leucopenia and immune system deficiency. The authors decided to present to colleagues their experience in this case, due to the presence of a rare heart abnormality and the operation technique for the treatment of this condition.

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