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Arthrogryposis and Abdominal Wall Defect Complicated With Intestinal Evantration in Pekingese Fetuses

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Abstract: A 3.5-year-old, pregnant Pekingese was presented with signs of dystocia. Vaginal examination revealed no fetuses in the birth canal, and no heart beats were observed in the fetuses, ultrasonographically. Four dead fetuses were retrieved by en bloc ovariohysterectomy. All fetuses had hind limb deformities and intestinal evantration was observed in three fetuses. A diagnosis of arthrogryposis and abdominal wall defect complicated with intestinal evantration was made after the macroscopic examination of the fetuses. The owner did not permit necropsy. To the best of our knowledge, this is the first report of arthrogryposis and abdominal wall defect complicated with intestinal eventration encountered in Pekingese fetuses.

Key Words: Abdominal wall defect, arthrogryposis, intestinal eventration, Pekingese fetus.

Pekinez Fetuslarında Artrogripozis ve İntestinal Evantrasyonla Komplike Abdominal Duvar Defekti

Özet: Gebe 3,5 yaşlı bir Pekinez güç doğum bulgularıyla getirildi. Vaginal muayene doğum kanalında fötus olmadığı açıkladı ve ultrasonografik muayenede fötuslarda kalp sesleri gözlenmedi. En-blok ovariyohisterktomi ile dört ölü fetus çıkarıldı. Tüm fötuslar arka ekstremite deformasyonuna sahipti ve üç fötusta intestinal evantrasyon gözlendi. Fötusların makroskopik muayeneleri sonrasında artrogripozis ve intestinal evantrasyonla komplike abdominal duvar defekti tanısı konuldu. Nekropsi hasta sahibi tarafından istenmedi. Bizim bilgimize gore Pekinez fetuslarında karşılaşılan artrogripozis ve intestinal evantrasyonla kompike abdominal duvar defekti ilk olgudur.

Anahtar Kelimeler: Abdominal duvar defekti, arthrogripozis, intestinal evantrasyon, Pekinez fötusu.

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Introduction

Congenital anomalies can be either functional or structural existing in newborns. These result from detrimental causes affecting during blastocyst, embryonal and fetal stages of the pregnancy. Although reports about the incidence of congenital defects are limited, their incidence is thought to be 1-2% in dogs[7].

Abdominal wall defect (AWD) is observed both in humans and animals[5,19,22,24]. This anomaly has several forms including umbilical hernia, gastroschisis, abdominoschisis and schistosomus reflexus (SR)[16]. Complete or partial protrusion of the abdominal organs from an abdominal fissure is called eventration[19,23]. Eventratio simplex is a protrusion of the intestines without envelope. If the protruded intestinal segments are enveloped by a membrane, it is called eventratio hernialis[3]. AWDs occur occasionally together with limb and vertebral defects[16,18,20]. Here, we describe arthrogryposis and AWD resulting in intestinal eventration in Pekingese fetuses and share these results with the veterinary practitioners.

Description of the Case

A 3.5 year-old, pregnant Pekingese was presented to Emergency Clinics of Faculty of Veterinary Medicine, Uludag University with signs of dystocia. Although the labor begun 9 hours ago and fetal fluids were discharged, no spontaneous delivery had occurred. The animal had had a caesarean operation at her previous pregnancy due to dystocia. Clinical examination revealed normal vital parameters, but a painful abdominal distention. Although cervix was dilated, there was no fetus or obstruction in the birth canal. No fetal heartbeat was detected ultrasonographically. These results were shared with the owner and *en bloc* ovariohysterectomy was decided.

After premedication and induction by xylazine HCl (1.5 mg/kg, im.) and ketamine HCl (10 mg/kg, im.) respectively, anesthesia was maintained with the combination of ketamine HCl and diazepam. The bitch was positioned ventrodorsally on the operation table and postumbilical midline incision was made after aseptic surgical preparation. There was no other abnormality in the abdominal cavity. *En bloc* ovariohyterectomy was performed with routine technique and incisions were closed.

Intramural examination of the uterus confirmed four dead fetuses. Macroscopically, three fetuses had an opening about 1 cm in diameter on the umbilical region and the intestinal segments evantrated from the umbilical opening (Figure 1). A membrane surrounding the intestines was not encountered. Furthermore, all fetuses had abnormal joint movement and axial deviations that was diagnosed as arthrogryposis in all fetuses. There was no other congenital deformity on the body surface. Unfortunately, the owner did not permit necropsy of the fetuses.

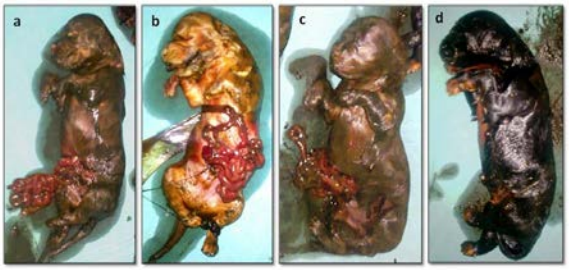

Figure 1. *This view demonstrates abdominal wall defects (approximately 1 cm diameter) on the umbilical region of three fetuses (**a, b** and **c**) except one (**d**).*

Şekil 1. *Bu resim biri hariç (**d**), üç fetusun (**a, b** ve **c**) umbilical bölgesindeki abdominal duvar defektini (yaklaşık 1 cm çaplı) göstermektedir.*

Discussion

It has been known that fetal anomalies cause dystocia[3]. The frequency of fetal malformation as a cause of dystocia has been reported as 1.6% in bitches[7]. We thought that fetal abnormalities were the cause of dystocia in this case.

Different types of AWD, including umbilical hernia, abdomino-thoracoschisis, omphalocel and gastroschisis have been described in several reports[1,16,18,21].

The AWDs are occasionally misdiagnosed as SR in clinic practice[5]. SR is a rare congenital defect and is a special form of abdominal fissures[12,15]. SR includes spinal inversion, exposure of abdominal viscera, limb ankylosis, abnormal positioning of the legs adjacent to the skull, lung and diaphragm hypoplasia that it is seen more commonly in cattle than other domestic animal species and humans[6,9,12,13,15-18]. Fissures in SR can be abdominal or thoracoabdominal[10,11]. Ozsoy *et al.* (2009) described thoracoabdominal fissure together with the dia-

phragm and heart anomalies in a dog. This fissure was not complicated with any spinal or limb deformity. They reported this condition as the first report of SR in dogs, which had similar macroscopic findings in calves defined by Aydin *et al*. (2006). In the presented case, we distinguished our case from SR because of the absence of vertebral deformities.

Although limb ankylosis and arthrogryposis are not typical forms for SR[2,3], perosomus elumbis (PE), another congenital anomaly, is characterized by lumbosacral vertebral and spinal cord agenesis, hind limb hypoplasia, arthrogryposis, limb ankylosis and muscular atrophy[2]. In present report, arthrogryposis was similar to PE, but limb hypoplasia was not observed.

Gastroschisis is described as fetal intestinal eventration from para-umbilical fissure covered with amniotic vesicle that it is commonly observed in humans[14]. In a report, gastroschisis has been described in a bovine embryo[1]. The presented case cannot be defined as gastroschisis, because the congenital deformity was observed on the umbilical region as abdominal wall defect complicated with intestinal eventration. Calzolari *et al*. (1993) described gastroschisis and omphalocel with limb deformity as "limb body wall complex" in humans. This case is compatible with this description; however, we did not observe such a definition in veterinary literature.

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