

Scientific Report

Congenital arthrogryposis associated with musculoskeletal defects in three newborn goats

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Summary

Arthrogryposis as a congenital malformation that refers to the fixed flexion of one or more joints as a result of stiffness or limited movements of multiple joints, change of posture and limb function due to permanent contracture of joints and wasting of muscles that usually occur at early stage of gestation. This report describes congenital arthrogryposis associated with musculoskeletal defects in three newborn goats. Both front and hindlimbs joints were characteristically flexed towards the anterior portion of the body. In addition, ankylosis, reduced muscle mass of limbs and rigidly fixed hindlimbs, lordosis, kyphosis and brachygnathia were observed. There was no definitive evidence of etiology of arthrogryposis, however, it seemed to be infectious, inherited or due to toxic plants. Since there is no recorded history about the mothers of the kids and the limited information available, causes of these defects cannot be determined.

Key words: Arthrogryposis, Lordosis, Kyphosis, Brachygnathia, Goat

Introduction

Congenital malformations with multiple anomalies have seldom been described in the veterinary literature. Arthrogryposis is described as a congenital joint involvement, non-progressive malformation from birth that usually affects several limbs alone or with others (Hall, 2013). Because of the complexity of the mechanisms resulting in the development of an anomaly, the precise causative agent of most congenital malformations is unknown (Kacar *et al.*, 2008). Arthrogryposis has more than one etiology and pathologic entity. Nevertheless, main etiologies are hereditary or genetic causes, although ingestion of toxic plants such as lupines and some viral infections during the early stages of gestation have also been suggested as the causative agents (Nawrot *et al.*, 1980). In Iran, some cases of congenital arthrogryposis have been reported in sheep and cattle (Khodakaram-Tafti *et al.*, 2000; Oryan *et al.*, 2011; Ozmaie and Aasghari, 2012) but there is no report about incidence and occurrence in goats as yet. Therefore, the present study reports the occurrence of congenital arthrogryposis associated with musculoskeletal malformations in three newborn goats.

Case presentation

Three newborn Iranian native goats (two male and one female) from two neighboring herds with 105 and 80

goats had the curvature with articular rigidity of joints in both forelimbs and hind limbs. There was five months between referrals of these kids. Method of husbandry of the herds was traditional (grazing in the pasture). Reduced muscle mass of forelimbs, extremely enlarged joints, angulation of hock joints rigidly fixed around the tibiotarsal joint were observed. One of the kids was born alive but unable to stand or nurse and died after 5 h. Slope of the back including lordosis or upward curvature of portion of the lumbar and spine associated with arthrogryposis was observed in this kid (Fig. 1). Arthrogryposis, brachygnathia, kyphosis or increased convex curvature of the spine and vertebral fusions were observed in two other kids. In addition, ankylosis, reduced muscle mass of limbs, rigidly fixed right hind limb, deviation of this hindlimb to the dorsal portion and in other hindlimb curvature of distal part to the outward were observed (Fig. 2). At necropsy, the internal organs were grossly normal in all affected kids. No other congenital anomalies such as cleft palate were seen. Gross and microscopic examinations of brains were normal in the kids.

Discussion

Congenital malformations are structural and functional abnormalities present at birth. They can affect a single structure or function, parts of various systems, or an entire system (Noden and De Lahunta, 1985).



Fig. 1: Arthrogryposis, reduced muscle mass of limbs, enlarged joints, angulation of hock joints and rigidly fixed around the tibiotarsal joint and lordosis are observed in a newborn goat



Fig. 2: Arthrogryposis associated with reduced muscle mass of limbs, rigidly fixed one of hindlimbs, deviation of this hindlimb to the back (thin arrow), curvature of distal part to outward in other hindlimb, brachygnathia (thick arrow) and kyphosis are seen in a kid

Arthrogryposis was first described in New Zealand in 1957. It literally means joint pie, that is defined as stiffness or limited movements of multiple joints and change of posture and limb function due to permanent contracture of joints at birth (Doherty *et al.*, 2000; Radostits *et al.*, 2007). The extent of this malformation is variable and may affect only one, two, or four legs and the axial skeleton (Van Vleet, 2007). Arthrogryposis usually affects the fore and hind limbs and the distal joints (Belli, 2007). This anomaly is reported more in calves, lambs, piglets and foals and less frequently in goats, cats, dogs and human beings (Doherty *et al.*, 2000; Schild *et al.*, 2002; Khodakaram-Tafti and Ikede, 2005; Radostits *et al.*, 2007; Devi Prasad *et al.*, 2010; Shahrokhi and Gharib, 2011). In general, the causative agents of arthrogryposis are most often unknown, but an accepted cause is probably reduced limb movement in

the uterus (Radostits *et al.*, 2007). Newborn animals affected with arthrogryposis are nearly always stillborn and may display autolysis due to intrauterine death 2-4 days early. Arthrogryposis may be associated with the other deformations such as palatoschisis or cleft palate, brachygnathia, scoliosis, lordosis, kyphosis, hydranencephaly and torticollis (Kacar *et al.*, 2008). In our report, arthrogryposis was combined with lordosis, brachygnathia and kyphosis. Severely affected calves are usually born dead and often cause dystocia, necessitating an embryotomy to deliver them (Leipold *et al.*, 1970). Arthrogryposis is not necessarily inherited as it may be due to environmental or infectious factors, however, it is known that many congenital disorders have a genetic cause (Greber *et al.*, 2013). Similar to our report, arthrogryposis has been characterized principally by symmetrical contracture of the four limbs (Greene *et al.*, 1973). Many of the affected animals have normal or smaller than normal skeletal size (Leipold *et al.*, 1974), but in our report the skeletal size of two fetuses were larger than normal. Arthrogryposis without central nervous system lesions is reported in ruminants born in herds where the gravid dams have been grazing. In the present study, no lesion was seen in central nervous system. Hereditary factors, teratogenic agents including ingestion of toxic and teratogenic plants by the dam, denervation in fetus and neonatal animals, vitamin A deficiency, selenium alone or with manganese deficiency as well as the administration of drugs like parabendazole and carbendazole are the etiologic possibilities (Nawrot *et al.*, 1980; Weisbrde, 2011). In cattle, the hereditary form of the disease has been proven in Shorthorn, Hereford, Jersey, and German Black Pied (Greene *et al.*, 1973, Nawrot *et al.*, 1980). The musculoskeletal system can also be affect by congenital neurologic disorders. Some toxic plants contain the piperidine alkaloids which produce a chemically induced reduction in fetal movements and may cause disorders in fetuses when the dam digests them during early pregnancy (Radostits *et al.*, 2007). The viral diseases such as bovine viral diarrhea, blue tongue, akabane disease, Cache Valley fever, Rift Valley fever, Aino virus and border disease can cause outbreaks of arthrogryposis in animals (Leipold *et al.*, 1974; Edwards *et al.*, 1989; Khodakaram-Tafti *et al.*, 2000; Belli, 2007; Devi Prasad *et al.*, 2010). Schmallenberg virus is a new emerging livestock disease that has been detected in cattle, sheep and goats and may cause severe birth defects in offspring born from infected animals such as arthrogryposis, torticollis, brachygnathia, scoliosis and hard muscles (DEFRA, 2011). However, the causes of most sporadic cases of arthrogryposis are undiagnosed, blue tongue virus commonly produces severe brain malformations in ruminants but has been shown to produce arthrogryposis only occasionally in calves. In outbreaks and epidemics, the damage to the nervous system associated with intrauterine infections or ingestion of plant toxins by the dam are potential pathogenesis (Weisbrde, 2011). In our report, gross and microscopic examinations of brain were normal in these fetuses. Oryan *et al.* (2011) reported

occurrence of arthrogryposis associated with cleft palate in the calves whose mothers were infected during early pregnancy with bovine viral diarrhoea virus or akabane virus infection or selenium and manganese deficiencies. Arthrogryposis by denervation or failure of innervation of skeletal muscle could result from intrauterine infection or toxin ingestion (Valentine, 2007). Hyperthermia may also induce occurrence of this deformation (Belli *et al.*, 2007). Congenital myodysplasia and arthrogryposis without any lesion in the brain was reported in sheep (Kacar *et al.*, 2008). In the uncommon cases, the neonate can be born alive if the limb involvement is unilateral (Schmidt and De Oliveira, 2004). In Iran, congenital arthrogryposis has been reported in sheep and cattle (Khodakaram-Tafti *et al.*, 2000; Oryan *et al.*, 2011; Ozmaie and Aasghari, 2012). With regard to existence of border disease virus in sheep herds of this region, complementary studies for diagnosis of definitive etiology of these defects are necessary.

References

- Belli, CB; Molina, LRP and Netto, DM (2007). A case report of arthrogryposis in caprine. *Ars. Vet. Jab. SP.*, 23: 67-70.
- DEFRA (Department for Environment, Food and Rural Affairs) (2011). Schmallenberg virus in northern Europe: a new orthobunyavirus in cattle. <http://www.defra.gov.uk/animal-diseases/a-z/schmallenberg-virus>.
- Devi Prasad, V; Hari Krishna, NVV; Sreenu, M and Thangadurai, R (2010). Arthrogryposis in a calf. *Vet. World*, 3: 335-336.
- Doherty, ML; Kelley, EP; Healy, AM; Callanan, JJ; Crosby, TF; Skelly, C and Boland, MP (2000). Congenital arthrogryposis: an inherited limb deformity in pedigree Suffolk lambs. *Vet. Rec.*, 146: 748-753.
- Edwards, JF; Livingston, CW; Chung, SI and Collisson, EC (1989). Ovine arthrogryposis and central nervous system malformations associated with in utero Cache Valley virus infection: spontaneous disease. *Vet. Pathol.*, 26: 33-39.
- Greber, D; Doherr, M; Drogemuller, C and Steiner, A (2013). Occurrence of congenital disorders in Swiss sheep. *Acta Vet. Scand.*, 55: 27-29.
- Greene, HJ; Leipold, HW and Huston, K (1973). Bovine congenital defects: arthrogryposis and associated defects in calves. *Am. J. Vet. Res.*, 34: 887-891.
- Hall, JG (2013). Uterine structural anomalies and arthrogryposis-death of an urban legend. *Am. J. Med. Genet. A.*, 161: 82-88.
- Kacar, C; Ozcan, K; Takci, I; Gurbulak, K; Ozen, H and Karaman, M (2008). Diprosopus, craniorachischisis, arthrogryposis and other associated anomalies in a stillborn lamb. *J. Vet. Sci.*, 9: 429-431.
- Khodakaram-Tafti, A and Ikede, BO (2005). A retrospective study of sporadic bovine abortions, stillbirth and neonatal abnormalities in Atlantic Canada, from 1990 to 2001. *Can. Vet. J.*, 46: 635-637.
- Khodakaram-Tafti, A; Kafi, M; Rouhani, K and Moahed, N (2000). Pathologic and serologic evidences of occurrence of border disease in sheep flocks of Fars Province of Iran. The 11th Iranian Veterinary Congress. 6-8 Mar., Tehran, Iran. PP: 366-368.
- Leipold, HW; Cates, WF; Radostits, OM and Howell, WE (1970). Arthrogryposis and associated defects in newborn calves. *Am. J. Vet. Res.*, 31: 1367-1374.
- Leipold, HW; Huston, K; Hulbert, LC; Guffy, M and Dennis, SM (1974). Congenital syndrome in Hereford calves with kyphoscoliosis, arthrogryposis and palatoschisis. *Cornell Vet.*, 64: 123-135.
- Nawrot, PS; Howell, WE and Leipold, HW (1980). Arthrogryposis: an inherited defect in newborn calves. *Aust. Vet. J.*, 56: 359-364.
- Noden, DM and De Lahunta, A (1985). *The embryology of domestic animals: developmental mechanisms and malformations*, Baltimore, Williams and Wilkins. PP: 109-152.
- Oryan, A; Shirian, S and Samadian, MR (2011). Congenital craniofacial and skeletal defects with arthrogryposis in two newborn male Holstein Friesian calves. *Comp. Clin. Pathol.*, 20: 43-46.
- Ozmaie, S and Aasghari, A (2012). A case of patent *Ductus arteriosus* and arthrogryposis in a buffalo calf. *J. Anim. Vet. Adv.*, 11: 2268-2269.
- Radostits, OM; Gay, CC; Hinchcliff, KW and Constable, PD (2007). *Veterinary medicine*. 10th Edn., Elsevier Science Health Science Division. PP: 1962-1978.
- Schild, AL; Soares, MP; Dame, MC; Portianski, EL and Riet-Correa, F (2002). Arthrogryposis in Murrah buffaloes in southern Brazil. *Pesq. Vet. Bras.*, 23: 13-16.
- Schmidt, V and De Oliveira, RT (2004). Arthrogryposis in a goat: a case report. *Arq. Bras. Med. Vet. Zootec.*, 56: 438-440.
- Shahrokhi, A and Gharib, M (2011). A case report of bilateral upper extremities arthrogryposis. *Ir. Reh. J.*, 9: 75-77.
- Valentine, BA (2007). Skeletal muscle. In: McGavin, MD and Zachary, JF (Eds.), *Pathologic basis of veterinary disease*. (4th Edn.), St. Louis, Mosby and Elsevier. P: 1002.
- Van Vleet, JF and Valentine, BA (2007). Muscle and tendon. In: Jubb, KVF; Kennedy, PC and Palmer, N (Eds.), *Pathology of domestic animals*. (5th Edn.), Vol. 1, University of Chicago, Elsevier Saunders, Academic Press Inc., PP: 204-206.
- Weisbrde, SE (2011). Bone and joints. In: McGavin, MD and Zachary, JF (Eds.), *Pathologic basis of veterinary disease*. (4th Edn.), St. Louis, Mosby Elsevier. PP: 1094-1095.