CASE REPORT

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Caudal duplication (*monocephalus tripus dibrachius*) in a kid goat

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SUMMARY

Dipygus is a congenital duplication in the caudal region of the body. According to the extension of the anomaly, duplicated cases are classified as *monoceophalus tripus dibrachius*, *monocephalus tetrapus dibrachius* and *cephalothoracopagus*. A newborn five-legged male kid was referred to the veterinary teaching hospital of Shahid Bahonar University of Kerman, Iran. In ventrodorsal radiography, normal pelvic symphysis was not observed between middle and left or right limbs. Necropsy findings included duplication of the bony pelvic girdle, alimentary tract from the jejunum downward, bladder and urethra. We hereby describe the first report of *monocephalus tripus dibrachius* in a kid goat.

Key words: Duplication – Monocephalus – Tripus – Dipygus – Kid goat

INTRODUCTION

Embryonic duplications as congenital abnormalities have been reported in small ruminants since 1916 (Dennis, 1975). It is believed that they result from errors happening during development (Noden and de Lahunta, 1985) and form a graded series from slight duplication to near-separation of two individuals (Hiraga and Dennis, 1993).

Dipygus is a kind of duplication in which the caudal region of the body has been affected and, according to the extent of the anomaly, has been classified as *monoceophalus tripus dibrachius, monocephalus tetrapus dibrachius* and *cephalothoracopagus* (Hiraga and Dennis, 1993). This anomaly has been reported frequently in cattle (Abt et al., 1962; Hiraga et al., 1989; Hiraga and Dennis, 1993; Leipold, 1972; Shojaei et al., 2010; Thakur, 1988) and sheep (Dennis, 1975; Doijode et al., 1992; Hiraga and Dennis, 1993), but not commonly in buffalos (Antoine et al., 1997; Thakur, 1988), and occurs very rarely in horses (Asquith and Sharp, 1979), dogs (Mazzullo et al., 2007; Nottidge et al., 2007) and cats (Seavers, 2009). As far as our knowledge goes, only three cases of similar anomalies in goats have been reported previously (Corbera et al., 2005; Mitra et al., 1994; Otiang'a-Owiti et al., 1997). In this paper, we present a case of dipygus which according to the available literature is the first description of *monocephalus tripus dibrachius* in a kid.

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Results

A newborn five-legged male kid was referred to the veterinary teaching hospital of Shahid Bahonar University of Kerman, Iran. The kid was very lethargic, and so was hospitalized, but the subject died a day after being born.

Before death, macroscopic examination showed three pelvic limbs at the pelvic region: left, middle and right (Fig. 1). The left limb seemed normal but the right one was rotated outwardly in a way that, distal to the femur, it was completely supinated. The middle limb was rotated slightly and attached to the body at the perineum.

Two anuses were present on both sides of proximal attachment of the middle limb, of which the right one was painful in palpation. A membranous duct which excreted urine continuously was seen under the right anus.

In radiographic evaluation, normal pelvic symphysis was not observed between either the left and middle or the middle and right limbs. In contrast with the left and right limbs which accommodated normally in the corresponding acetabulums, the articulation of the middle limb with a normal acetabulum was not seen.

Necropsy revealed that duplication occurred in the internal organs as well as the bony pelvic girdle. Four sets of hip bones, positioned sequentially from left to right (numbered 1-4 respectively), were seen (Figure 2). Hip bones 1 and 4 were almost formed normally. The hip bone number 1 and a vestige of the ischiatic part of hip number 2 had made only the ischiatic part of left symphysis. Besides right symphysis, hips number 3 and 4 fused to each other at sacral tubers. Hips number 1 and 4 had normal acetabula that accommodated the head of the femurs of left and right pelvic limbs, respectively. Distal to the iliac wings of the 2nd and 3rd hip, two bones were fused to form a single body and a common acetabulum for the middle limb. Two wings of the aforementioned hips were incompletely separated from each other by a groove. A common pubis and ischium with an obturator foramen was distinguished near the common acetabulum which, by the hip number 4, formed the right symphysis.

Regarding the features of bone components, the middle limb was potentially a left

Fig. 1. Three pelvic limbs in the affected kid goat.

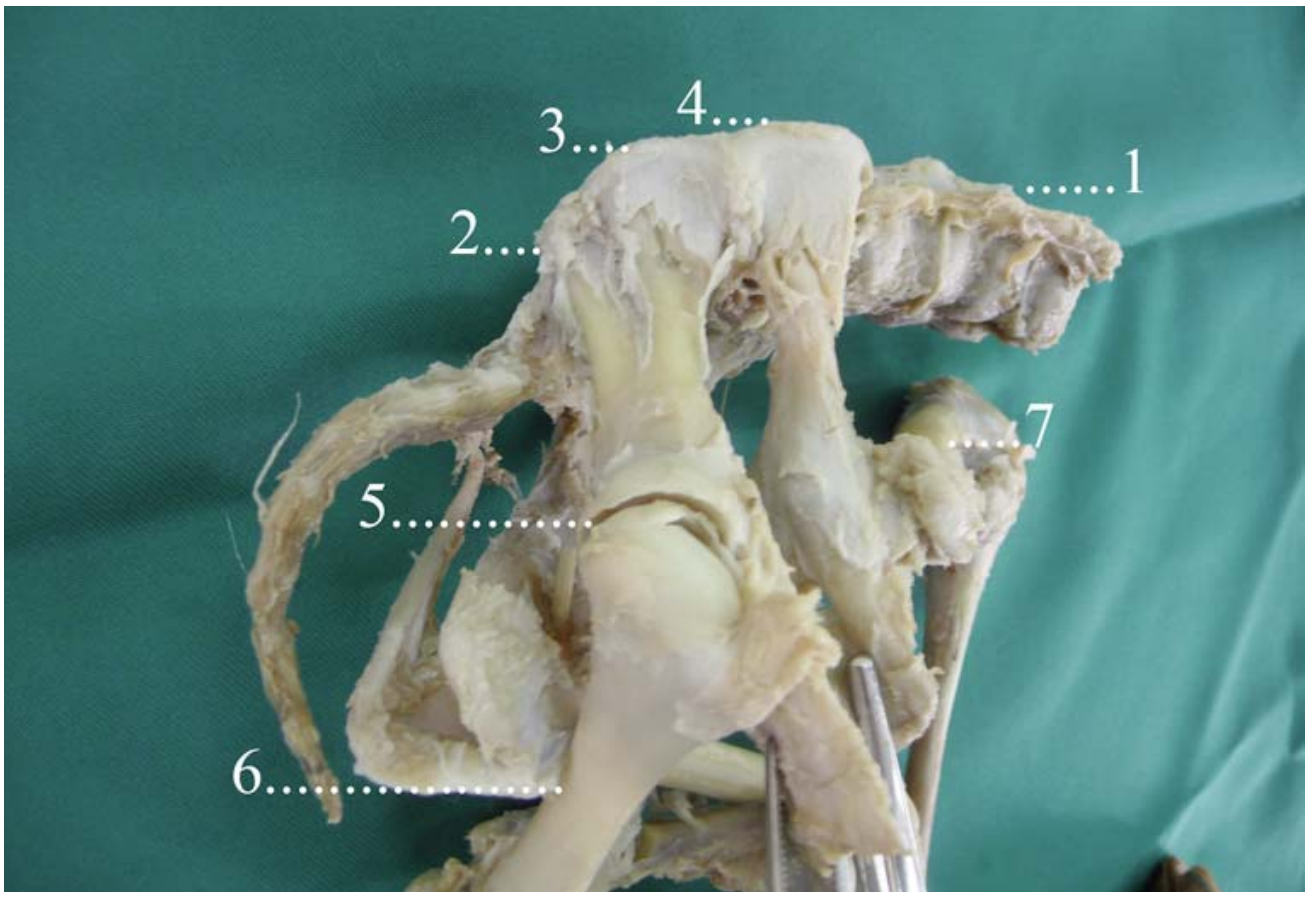

Fig. 2. Hip bones number 2 to 4. 1: Vertebral column, 2: Ilium of hip bone number 2, 3: Ilium of hip bone number 4, 5: Common acetabulum of hip bones 2 and 3, 6: Middle femur, 7: Head of right femur.

limb with complete bony elements of a normal limb.

Spina bifida was present at the sacral region. The sacrum, with a small deviation to the right side, was seen between hip bones number 1 and 2.

The digestive system was formed normally up to the distal part of the jejunum, beyond which the gut was duplicated distally to form two complete sets of ileum, cecum, colon, rectum and anus. The left anus was opened in the normal position (ventral to the tail); however, the right one was observed between the middle and right limbs.

Two kidneys were seen in their normal positions. The left and right ureters entered to the necks of the dorsal and ventral urinary bladders respectively. The dorsal bladder was full of urine but the ventral one was empty (Figure 3).

Each bladder was continued by an urachus cranially and a urethra caudally. The dorsal urethra had entered the crus of the penis as the extra-pelvic part in the perineal region and coursed to the tip of the penis. Ventral urethra had extended and opened between the right and middle limbs as a membranous duct.

The penis was developed in its normal position. The dorsal and ventral urethras had received the left and right deferent canals respectively. The right testis was seen in the abdominal cavity, over the vaginal rings and the left one in the inguinal canal, near the scrotum. Moreover, two small testicular structures were observed near the kidneys.

Two caudal mesenteric arteries were seen sequentially, the cranial and caudal which had furnished the right and left hind guts respectively. Two umbilical arteries were seen on the right side and between two bladders. They had branched off from two internal iliac arteries. Three hind limbs were furnished by three external iliac arteries which had directly branched off from the aorta.

Discussion

Conjoined twinning in humans is expected to occur if the embryonic disc divides later than the 13th day post fertilization, as this

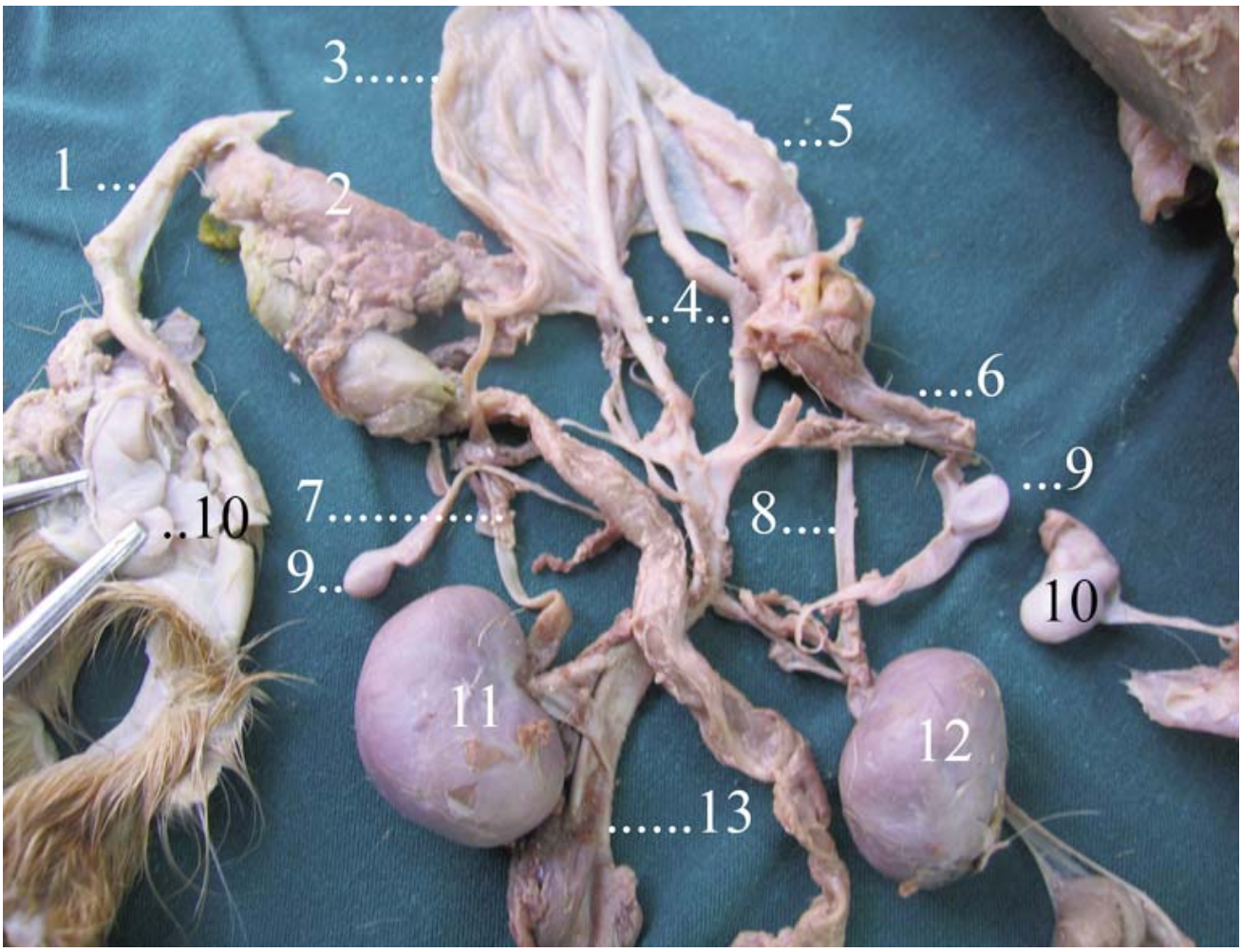

Fig. 3. Affected urogenital system. 1: Penis, 2: Dorsal urethra, 3: Dorsal urinary bladder, 4: Umbilical arteries, 5: Ventral urinary bladder, 6: Ventral urethra, 7: Left ureter, 8: Right ureter, 9: Testicle-like structures, 10: Main testes, 11: Left Kidney, 12: Right Kidney, 13: Aorta.

may result in an incomplete division (Sarihan et al., 1995). There is no data available on the precise chronology of this anomaly during the pregnancy in ruminants, but it is generally believed that, if twinning is not initiated until after the embryonic disc is formed, two centers of axial growth may result instead of one. So, if they are not sufficiently separated, conjoined twins could result (Hiraga and Dennis, 1993). According to the specific developmental features of this case, the initial developmental abuse might have affected the embryo between 13.5 and 21 days postovulation which includes the primitive streak stage in the goat (Molinari and Goicoechea, 1993 in Otiang'a-Owiti et al., 1997).

Congenital involvement of caudal duplication in sheep compared with the cranial one in cattle has been reported (Dennis, 1975), but according to the few reports of duplication in goats (Corbera et al., 2005; Mitra et al., 1994; Mukaratirwa and Sayi, 2006; Otiang'a-Owiti et al., 1997; Ramadan, 1996) it cannot be concluded that the occurrence of this anomaly is more often cranial or caudal.

In the literature review, we can only find three cases of dipygus in goats; one with eight (Mitra et al., 1994), and the other two with six legs (Corbera et al., 2005; Otiang'a-Owiti et al., 1997) which have been classified as *cephalothoracopagus* and *monocephalus tetrapus dibrachius*, respectively (Hiraga and Dennis, 1993). Although Otiang'a-Owiti et al. (1997) reported their case as parasitic attachment of an extra set of pelvic limbs, the duplication of internal organs in that case is very similar to ours. Duplication of the gastrointestinal tract from the mid jujenum downward, two urinary bladders which (each receiving a ureter from a respective kidney), duplicated urethra and unduplicated right and left paramesonephric (Otiang'a-Owiti et al., 1997), or mesonephric (present case) ducts which form the genital duct of each twin, are striking similarities of these two cases. Two other similar dipygus cases were found in humans with the duplica-

tion of the ileum downward, duplication of the bladder, urethra and genitalia, and the abnormal curvature of sacrum and/or its duplication (Bannykh et al., 2001; Kroes et al., 2002). These latter findings have been mentioned in human as "caudal duplication syndrome" (Dominguez et al., 1993).

Conjoined twins most frequently result from a single embryonic disc that has undergone splitting between a single dorsalward amniotic cavity and a single ventralward yolk sac (Machin, 1993). Although the exact mechanism(s) is not clear, the most readily accepted theory is based on the embryonic fission and formation of two organizing centers, such as two primitive streaks and/or notochordal axes (Spencer, 1992; Machin, 1993; Dominguez et al., 1993; Hiraga and Dennis, 1993). Noticeable similarities of duplicated organs and unduplicated structures which originate from the cranial parts of the embryonic disc and are subsequently situated in the pelvic region (mesonephric and paramesonephric ducts) (Otiang'a-Owitiet al., 1997; Bannykh et al., 2001; Kroes et al., 2002; present case) are evidence for those who believe that, in "caudal duplication syndrome", teratogen affects the embryo during the disc formation and possibly by a similar process. Unduplicated kidneys which are seen in all cases may be due to their unduplicated inducers (mesonephric ducts) which originate from the cranial parts of the embryo.

Duplication of the urethra and genitalia, as associated defects in the dipygus cases, have been frequently observed (Abt et al., 1962; Anastasakis et al., 2007; Bannykh et al., 2001; Hiraga et al., 1989; Kroes et al., 2002; Leipold et al., 1972; Mazzullo et al., 2007; Rattan et al., 2000; Thakur, 1988), but a duplicated bladder accompanied by duplicated urethra and genitalia has been rarely reported in humans (Bannykh et al., 2001; Kroes et al., 2002; Ulman et al., 1996) or animals (Antoine et al., 1997; Mazzullo et al., 2007; Otiang'a-Owiti et al., 1997).

Although several sources have reported the occurrence of Dipygus with three hind legs in humans (Anastasakis et al., 2007; Chao et al., 1980; Doski et al., 1997; Holcomb, 1989; Spitz et al., 1997; Yokomori et al., 1993), there are very few reports about this anomaly in animals (Abt, 1962; Mazzullo et al., 2007; Thakur, 1988). We believe that this is the first description of the *monocephalus tripus dibrachius* in a kid.

Etiological information about the congenital duplication anomalies is rarely available. It is not known precisely whether they are caused by genetic or environmental factors, or both (Hiraga and Dennis, 1993). There are reports of cranial or caudal duplications in the Kerman province of Iran (Shojaei et al., 2006; Shojaei and Asadi, 2008; Shojaei et al., 2010; two unpublished cases of the *cephalothoracopagus* and *thoracopagus* in goats). However, due to the climate variation and the seasonal movement of the animals, investigation of the environmental causes of the anomaly is very difficult. In our case, there was no history of any similar anomaly in the herd, nor of specific teratogen and/or drug administration.

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References

Abt DA, Croshaw JE, Hare WC (1962) Monocephalus dipygus parasiticus and other anomalies in a calf. *J Am Vet Med Assoc*, 141: 1068-1072.

Anastasakis E, Zhang EG, Bates AW, Abdel-Aal MAE, Kadir RA (2007) Parapagus dicephalus dibrachius tripus: an unusual case of conjoined twins. *Prenat Diagn,* 27: 1165-1166.

Antoine D, Murugavel K, Alphonse RMD, Ramalingam S, Thandavamurthy C (1997) Monocephalus dipygus monster in a buffalo – a case report. *Ind J Anim Reprod,* 18: 166.

Asquith RL, Sharp DC (1979) An equine fetal monster dicephalus tetrapus dibrachius. *Theriogenology,* 12: 39-44.

Bannykh SI, Bannykh GI, Mannino FL, Jones KL, Hansen L, Benirschke-Masliah KE (2001) Partial caudal duplication in a newborn associated with meningomyelocele and complex heart anomaly. *Teratology,* 63: 94-99.

Chao CC, Susetio L, Luu KW, Kwan WF (1980) Anaesthetic management for successful separation of tripus ischiopagal conjoined male twins. *Can Anaesth Soc J,* 27: 565-571.

Corbera JA, Arencibia A, Morales I, Gutierrez C (2005) Congenital duplication of the caudal region (*monocephalus dipygus*) in a kid goat. *Anat Histol Embryol,* 34: 61-63.

Dennis SM (1975) Embryonic duplications in sheep. *Aust Vet J,* 51: 83-87.

Doijode SV, Thorat NH, Markandeya NM, Mamde CS (1992) Monocephalus dipygus conjoined twin lamb. *Ind J Anim Reprod,* 13: 206.

Dominguez R, Rott J, Castillo M, Pittaluga RR, Corriere JN (1993) Caudal duplication syndrome. *Am J Dis Child,* 147: 1048-1052.

Doski JJ, Heiman HS, Solenberger RI, Stefko RM, Kuivila T, Rozanski TA, Flack CA, Thurman RT, Milbourn CT, Cheu HW (1997) Successful separation of ischiopagus tripus conjoined twins with comparative analysis of methods for abdominal wall closure and use of the tripus limb. *J Pediatr Surg,* 32: 1761-1766.

Hiraga T, Dennis SM (1993) Congenital duplication. In: Dennis SM (Ed). *Congenital abnormalities*. *Vet Clin North Am,* 9: 145-161.

Hiraga T, Abe M, Iwasa K, Takehana K, Tetsuka M (1989) Seven-legged calf-dipygus with an extra foreleg at the pelvic region. *Nippon Juigaku Zasshi,* 51: 1011-1015.

Holcomb GW (1989) Continent urinary reconstruction in ischiopagus tripus conjoined twins. *J Pediatr Surg,* 24: 721-731.

Kroes HY, Takahashi M, Zijlstra RJ, Baert JALL, Kooi KA, Hofstra RMW, van Essen AJ (2002) Two cases of the caudal duplication anomaly including a discordant monozygotic twin. *Am J Med Genet,* 112: 390-393.

Leipold HW, Dennis SM, Huston K (1972) Embryonic duplications in cattle. *Cornell Vet,* 62: 572-580.

Machin GA (1993) Conjoined twins: Implications for blastogenesis. Birth Defects: Original Article Series, 29: 141-179.

Mazzullo G, Monteverde V, Macri F, Partanna S, Caracappa S (2007) Incomplete caudal duplication in a puppy: gross and radiological observations. *J Small Anim Pract,* 48: 410-413.

Mitra M, Basak DN, Chakrabarti A (1994) Monocephalus thoracopagus tetrapus tetranrachius monster in a Black Bengal goat. *Ind Vet J,* 71: 177-178.

Mukaratirwa S, Sayi ST (2006) Partial facial duplication (diprosopus) in a goat kid. *J S Afr Vet Assoc,* 77: 42-44.

Nottidge HO, Omobowale TO, Olopade JO, Oladiran OO, Ajala OO (2007) A case of craniothoracopagus (monocephalus thoracopagus tetrabrachius) in a dog. *Anat Histol Embryol,* 36: 179-181.

Otiang'a-Owiti GE, Oduor-Okelo D, Kamau GK, Makori N, Hendrickx AG (1997) Morphology of a six-legged goat with duplication of the intestinal, lower urinary, and genital tracts. *Anat Rec,* 247: 432-438.

Ramadan RO (1996) A dicephalic goat with other defects. *Zentralbl Veterinarmed A,* 43: 337-343.

Rattan KN, Budhiraja S, Pandit SK, Solanki RS, Sen R, Sen J (2000) Caudal duplication - a case report. *Pediatr Surg Int,* 16: 445-446.

Sarihan H, Mocan H, Aslan M, Akyazici R (1994) A rare case of truncal duplication. *Pediatr Surg Int,* 29: 1517-1518.

Seavers AM (2009) Monocephalus dipygus parapagus: a suspected case of complete caudal duplication in a British Blue kitten. *J Feline Med Surg,* 11: 330-331.

Shojaei B, Asadi A (2008) Dicephalus, inferior brachygnathia, cleft palate and campylognathia in a calf. *Iran Vet J,* 4: 117-122.

Shojaei B, Derakhshanfar A, Oloumi MM, Hashemnia S (2006) Diprosopus, spina bifida and kyphoscoliosis in a lamb. *Vet Arhiv,* 76: 461-469.

Shojaei B, Kheirandish R, Hasanzade M (2010) Monocephalus tetrapus dibrachius in a calf. *Comp Clin Pathol,* 19: 511-513.

Spencer, R (1992) Conjoined twins: theoretical embryologic basis. *Teratology*, 45: 591-602.

Spitz L, Capps SNJ, Kiely EM (1991) Xiphoomphaloischiopagus tripus conjoined twins: Successful separation following abdominal wall expansion. *Pediatr Surg Int,* 26: 26-29.

Thakur SB (1988) Dipygus monstrosity in a calf. *Ind Vet J,* 65: 440.

Ulman I, Avanoglu A, Erdener A, Sahin AH, Gokdemir A (1996) Evaluation of the lower urinary tract function in caudal duplication (dipygus) anomaly. *J Pediatr Surg*, 31: 1680-1681.

Yokomori K, Ohkura M, Kitano Y, Nakajo T, Harii K, Tanikaze S (1993) Comprehensive planning of operative strategy for separation of ischiopagus tripus twins with particular reference to quality of life. *J Pediatr Surg,* 28: 833-837.