THE ANATOMICAL RECORD 247:432–438 (1997)

# Morphology of a Six-Legged Goat With Duplication of the Intestinal, Lower Urinary, and Genital Tracts

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***ABSTRACT*** ***Background:*** **An adult female goat with rare malformations, which consisted of duplication of the intestinal, lower urinary, and genital tracts as well a pair of parasitic appendages, is presented.**

***Methods:*** **A complete dissection was performed on a moribund female goat (*Capra hircus*).**

***Results:*** **The animal had a normal body with a parasitic attachment located within the pelvic region. This attachment was represented by an ovoid, trunk-like, adipose mass that lacked internal organs or vertebrae but that had two fairly well-developed limbs with the normal components of hind limbs. There was duplication involving the external and internal genitalia, the urethra, the urinary bladder, and portions of the small intestine as well as the large bowel, including the anal openings.**

***Conclusions:*** **An autosite with a duplication involving the hindgut and paramesonephric anlages was identified. These features were compatible with life in utero and postutero and emanated from incomplete twinning (heteropagus twins). A review of the literature also suggests that heteropagus twins are a very rare abnormality in both domestic animals and humans. Anat. Rec. 247:432–438, 1997** 

**Key words: heteropagus; parasitic conjoined twins; intestinal duplication; bladder duplication; genital tract duplication; goat anomaly; anatomy**

The presence or birth of conjoined twins and other related forms of anatomical duplications have always fascinated lay persons and clinicians alike. They represent a sequel of embryonic duplications that result from the splitting or abnormal duplication of embryonic structures during early stages of development. The case report discussed here was presented as a nonviable specimen with a normal-looking body, two anal openings, two sets of external genitalia, and an attached parasitic twin represented by a caudoventral mass with two limbs. Such anomalies fall under the category of parasitic, heteropagus, or asymmetrical conjoined twins (Guttmacher and Nicholls, 1967; Potter and Craig, 1975). In these cases, one member, the "host" (or autosite), is anatomically normal and bears an incomplete cotwin, the "parasite," whose growth and survival is solely dependent on the host. The asymmetrical or parasitic conjoined twins (PCTs) form a subset of congenital duplications, the better-known subset being the conjoined, symmetrical, diplopagus or "Siamese" twins, in which each member is equal and symmetrical with its partner; their classification is normally based on the degree of fusion and the most prominent site of conjunction between the twins. Information on the definitive etiopathological factors of these duplications still remains obscure, but it is now known that they are a complicated variety of monozygotic twinning, which is a product of a single fertilized ovum (Arey, 1965; Hamilton et al., 1966).

The precise incidence of conjoined twins in domestic animals and man has not been fully determined, most likely due to high prenatal mortality as well as incomplete or poor documentation. It is estimated that these cases occur in about 1 in 100,000 births in man (Harper et al., 1980; Edmonds and Layde, 1982). Although the incidence is equally low in farm animals, these malformations are most common in cattle (Arthur, 1956; Leipold et al., 1972; Greene et al., 1973; Roberts, 1986; Szabo, 1989), less frequent in sheep (Dennis, 1975) and pigs (Selby et al., 1973; Partlow et al., 1993), and extremely rare in horses (Roberts, 1986) and goats (Basrur, 1993). Conjoined symmetrical twins account for a considerably larger percentage of the twinning reported in both cattle and pigs when compared with other types of twinning (Arthur, 1956; Leipold et al.,

Received 10 March 1996; accepted 12 September 1996.
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1972; Selby et al., 1973; Roberts, 1986; Hiraga and Dennis, 1993). The PCTs are least common in these species and appear to be the more common form of twinning observed in sheep (Leipold et al., 1972; Dennis, 1975; Hiraga et al., 1989). Only two cases of conjoined twins have been documented to date in goats (Basrur, 1993).

Although PCTs with various forms of anatomical duplications have been described in cattle (Abt et al., 1962; Leipold et al., 1972; Hiraga et al., 1989), sheep (Dennis, 1975), and pigs (Selby et al., 1973; Partlow et al., 1993), a review of the literature indicates that there is only one general description of a parasitic asymmetrical female calf twin with a duplication of the vagina and anus comparable to the one in the present study (Leipold et al., 1972). A few cases of PCTs in humans that lived beyond childhood have been documented or mentioned (Stephens et al., 1982; Sondrarajan et al., 1994). Furthermore, there are no anatomical descriptions of PCTs in domestic animals that have lived beyond sexual maturity. This inadequacy is most likely due to the fact that most PCTs either are usually stillborn, die soon after birth, or are of no economic value to the farmer. Due to the unique features identified, this case is presented because it represents the first anatomical description of a goat PCT that had lived beyond sexual maturity. This report also elucidates the possible embryological events that may have led to the development of these anomalies.

## MATERIALS AND METHODS

A 30 kg adult female goat (*Capra hircus*) of roughly 4 years of age was presented to the Department of Veterinary Anatomy, University of Nairobi, Kenya, from the large animal hospital following its death. This animal had been brought to the hospital earlier exhibiting tetanus-like symptoms, according to the clinician's report. It died prior to admission, and the cause of death was ascribed to septicemia. No postmortem was done. The early postnatal history was sketchy apart from the fact that there was no history of congenital anomalies in the flock. The owner claimed that this animal had previously given birth to normal offspring. He had earned a good living displaying this animal in various Agricultural exhibitions within the country.

After a thorough external inspection and photographic documentation of all recognizable malformations, the animal was entirely immersed in 10% formalin, because attempts at perfusion through the vascular system were unsuccessful due to already-established intravascular coagulation. Radiography capabilities were not available for complete skeleton evaluation at the time of study, and, following fixation, an anatomical dissection was performed.

## RESULTS

The gross external features of the autosite were essentially normal from the head up to the perineum and pelvis (Fig. 1). Within the region of the pelvic and perineum, there was a parasitic twin, which was directed ventrally between the two normal hind limbs (Figs. 2, 3). The external form of this caudoventrally located parasite consisted of an ovoid mass that resembled the lower part of the trunk with two fairly


Fig. 1. Lateral view of the autosite showing the general external appearance. Note the two extra limbs cranial to the two normal hind limbs.


Fig. 2. A ventral view of the autosite showing the position of the parasitic twin and its limb appendages.

well-developed limbs that were shorter than the normal autosite limbs. These limbs were flexed, were directed cranially, and responded positively to induced flexure. Each possessed normal components (skeleton and muscular) of the hind limbs. From their appearance, it may be speculated that these limbs were occasionally utilized to support the body weight of the autosite when it was changing its posture, for example, from a sitting to a standing position, and vice versa. Because the cotwin occupied parts of the inguinal area, the two mammary glands of the host were positioned slightly cranially to occupy the area between the ventral abdominal wall and the lower wall of the parasite. There was no tail, perineum, or any openings on the surface of the parasite. The parasitic skin was continuous with that of the autosite at all points of attachment.

Dissection of the trunk-like mass of the parasite revealed a mass of adipose tissue with neither vertebral structures nor visceral organs. A few small nerve bundles were evident subcutaneously. The two limbs


Fig. 3. A rear view of the autosite and its parasitic attachment showing their relationship.

were not fused to each other but were entirely separate. Careful dissection revealed that each limb articulated with the ventral surface of the ischium (left and right) through a moveable joint with a joint capsule. The autosite and the twin appendages did not share organs or major blood vessels, and the cotwin appeared to be nourished by cutaneous vasculature and two small vessels that originated from the host's perineal artery.

Dorsal to the parasitic twin, there were two anal openings and two sets of external genitalia (Fig. 3, 4). When viewed from the caudal aspect, the two anal openings were separate from each other and were symmetrically placed on either side of the perineal midline, in line with their corresponding external genitalia. Judging from their appearance and the presence of fecal staining, each orifice appeared to be functional and most likely had discharged feces. The gastrointestinal tract was single and was relatively normal up to the level of midjejunum, where it bifurcated (Fig. 5). From this point onward, the remainder of the intestinal tract was doubled or duplicated. All of the components of a normal intestinal tract at this level were evident, with mirror-image jejunum, ileum, cecum, colon, rectum, and anal openings. The arrangement within each of the two duplicated portions was identical to that normally seen in a dissection of an adult goat. The junction of the jejunum and ileum was defined by the ileocecal fold; the spiral loops (centrifugal and centripetal coils) of the ascending colon as well as the transverse and descending colon were evident. The two large bowels were of normal size and shape and were completely separate. They measured roughly 240 and 230 cm, respectively, from the point of bifurcation to the anus. Both tracts were supported and surrounded by a single extensive mesentery, which had to be severed at several points in order to display the duplicated intestinal loops. There was a single cranial mesenteric artery, but, in the area of the duplication, this vessel divided into numerous smaller branches, and it was difficult to ascertain the existence of a dual mesenteric supply to this region. Intestinal and fecal contents were present in each portion up to the anus, a fact that further confirmed our earlier observation regarding functionality.

The two normal-appearing vulvae were widely separated from the midline and were located more toward

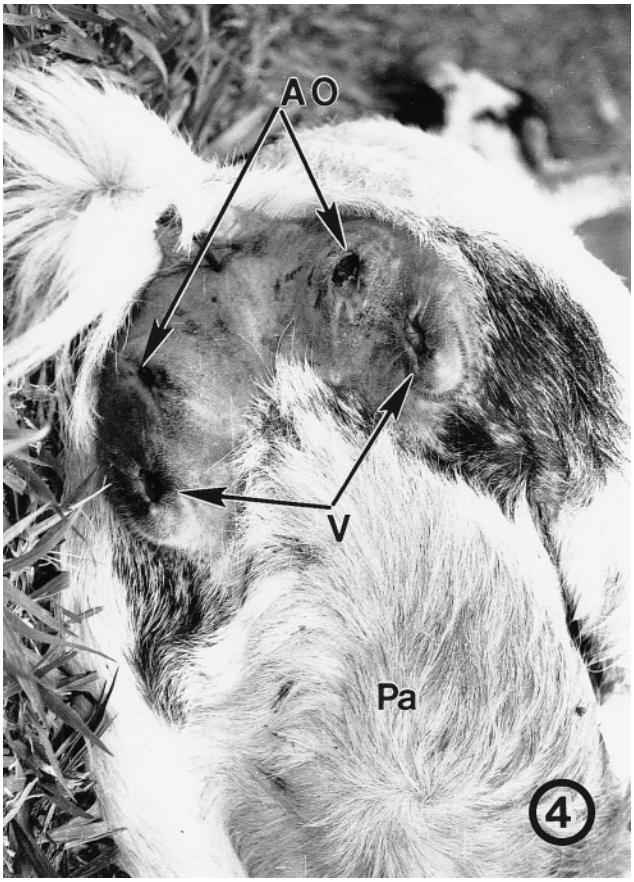


Fig. 4. A rear view of the autosite's perineal region demonstrating the two anal openings (AO), two vulvae (V), and the trunk-like mass of the parasite (Pa).

the lateral aspect of the perineum (Fig. 4). Internally, each vulva led to separated internal genitalia that were mirror images of one another (Fig. 5). Each genital tract had a vestibule, clitoris, vagina, cervix, unicornous uterus, and an ovary. Furthermore, each tract was suspended from the body wall by a single fold of peritoneum that represented part of the broad ligament. Judging from the condition of the tracts (size and presence of caruncles within each horn) and ovaries (presence of a large corpus luteum on the right ovary and several corpus albicans on the left), it was apparent that this autosite was reproductively sound, confirming the owner's claim regarding parity. There were two completely separate urinary bladders, each firmly anchored by connective tissue to the ventral surface of the vagina. Each received a single ureter and emptied into individual urethra located on the floor of the vestibule. Both urinary bladders were of relatively normal size with an apex, body, and neck. The autosite had two kidneys of normal size, and each emptied into the ipsilateral bladder via a single ureter. Each tract within the pelvic viscera of the autosite had its own fold of tissue that suspended the viscera from the body wall. In addition, there were no connections between these pelvic viscera and any tissues of the twin appendage.

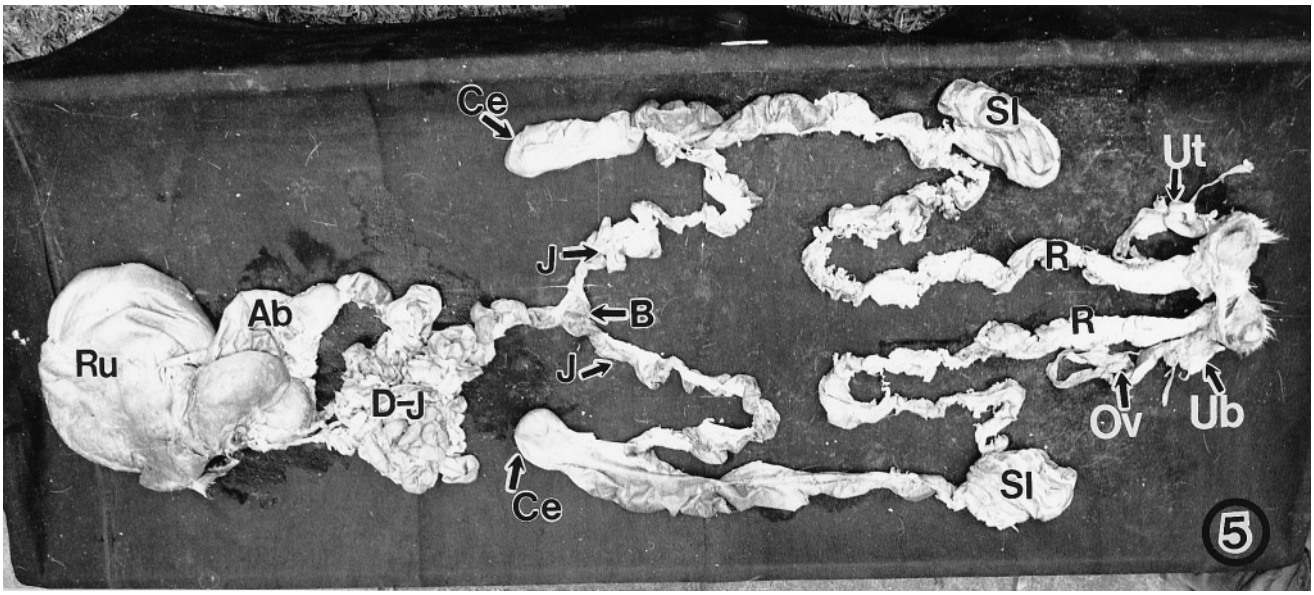


Fig. 5. The displayed gastrointestinal and urogenital tracts from the autosite. Note the point of intestinal bifurcation (arrow with B). Ab, abomasum; Ce, cecum; D-J, duodenum and portions of jejunum; J, jejunum; Ov, ovary; R, rectum; Ru, rumen; Sl, spiral loop of ascending colon; Ub, urinary bladder; Ut, uterine horn.

Finally, no notable anomalies were discernible in the head, neck, or thoracic viscera of the autosite.

## DISCUSSION

The PCT presented in this study can be classified either as a monocephalus tetrapus dibrachius (Guttmacher and Nicholls, 1967; Potter and Craig, 1975) or as an ischiopagus parasiticus (Nishimura and Okamoto, 1976). PCTs are rare duplications in both domestic animals and man. In these cases, the parasite is normally visible on the external surface of the autosite and may be randomly oriented on any surface (epigastrium, head, sacrum, or pelvis). Many varieties of PCTs have been described in both domestic animals (Goss and Cole, 1945; Bhattacharyya, 1964; Leipold et al., 1972; Dennis, 1975; Selby et al., 1973; Hiraga et al., 1989; Hiraga and Dennis, 1993) and man (Rowe et al., 1968; Simpson et al., 1973; Potter and Craig, 1975; Nishimura and Okamoto, 1976; Spitz et al., 1979; Stephens et al., 1982; Husain et al., 1989; Takayanagi, 1991; Drut et al., 1992; Chadha et al., 1993a,b); however, the majority of these cases actually represent duplication of the caudal region (dipygus) with an attachment of a parasitic or rudimentary pelvis and one or two rudimentary limbs in and around the perineum or pelvis of the autosite, similar to the findings reported here.

The duplication of the various portions of the urogenital and lower intestinal tracts with or without parasitic attachments are extremely rare abnormalities in both domestic animals (Leipold et al., 1972; Dennis, 1975; Hiraga et al., 1989; Longhofer et al., 1991) and man (Ravitch, 1953; Beach et al., 1961; Breen and Weinberg, 1965; Rowe et al., 1968; Smith, 1969; Veeraraghavan et al., 1983; Zamir et al., 1984: Magbool et al., 1988; Okur et al., 1992). The present case involved the combination of duplicated portions of the intestinal, lower urinary, and genital tracts with an accompanied parasitic appendage in the pelvic region; such situations are extremely rare into adulthood. Among the domestic animals, only Leipold et al. (1972) noted a viable female calf with similar features. Although this case included four normal limbs, two accessory hind limbs attached to the normal pelvis, and duplicated anus and vagina, like many other reports on PCTs, the observations addressed the external gross features only. The external morphological appearance of the present case resembles that shown for a lamb (depicted in Fig. 17 of Hiraga and Dennis, 1993) and those described in some duplicitas posterior mice (Center, 1969). Furthermore, this case had several manifestations of the features described in human infants by Rowe et al. (1968) and Simpson et al. (1973), where two sets of external genitalia, two anal openings, and four lower extremities were encountered. Similar to the goat reported here, the external genitalia and anal openings were within the autosite itself in both these cases. These situations are unlike those observed in most instances of PCTs, where the urogenital and intestinal tracts are usually bilaterally expressed in both twins.

It is well known that conjoined twins most frequently develop 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 mechanism(s) involved in these cases is largely unknown, 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). It is further suggested that, once they are formed, these

structures are initially destined to form two separate individuals. But they grow in close proximity, coalesce, and, subsequently, development becomes centered around these axes (Spencer, 1992). The variation in the orientation and separation of the axes in addition to the subsequent development of individual organs ("neo-axes") presumably accounts for the different types of conjoined twins that can occur.

Little is known regarding the pathogenesis of PCTs. Similar to other forms of conjoined twinning, they seem to represent severe aberration of the normal twinning processes. It has been suggested that PCTs, together with dipygus conjoined twins, represent residua of previously more extensive conjoined twins that have undergone subsequent disruptive and degenerative process in only one axes (Machin, 1993). Although an individual case description is not in itself sufficient to explain the pathogenic sequence in the case of a congenital anomaly, a few suggestions are presented regarding the pathogenesis of the defects noted in this case. It is clear that the anomalies in the present case were a manifestation of a disturbance in the development of the caudal end of the embryo. On the basis of the specific developmental criteria noted, it is suggested that the initial developmental insult might have occurred between 13.5 and 21 days postovulation. This period encompasses the primitive streak stage in the goat (Molinari and Goicoechea, 1993). Using the theories adopted from Spencer (1992) and Machin (1993), we suggest that the dipygus-equivalent appearance evident in our case by the presence of two sets of external genitalia, two anuses, and four hind limbs emanated from an attempted twinning. This was initiated either by a singular early embryonic split/duplication or by two separate embryonic organizers at the caudal end of the embryo. However, during subsequent embryogenesis in either situation, for unknown reasons, one axis may have failed to develop properly, and the resulting twin subsequently became assimilated in or attached to the autosite.

The parasitic attachment consisted of a pair of limbs and an adipose mass, with an apparent competition between the parasitic pair and the primary limbs during development. Based on the relationship of the attachment with the pelvis, we strongly believe that the supernumerary limbs emanated from the failure of the twinning process. However, it should be noted that a coincidental development of duplicate limb structures possibly can occur from abnormalities of the apical ectodermal or Wolffian ridge during early limb morphogenesis, a situation that has been considered in cases of limb duplication (Goetinck, 1964; Sledge, 1966; Stephens et al., 1982).

Axial structures and visceral organs were conspicuously absent within the parasitic twin, a situation that is difficult to explain. In most cases of parasitic twins, there are typically remnants of either vertebral structures or visceral organs. One possible explanation is based on the developmental concept by Machin and Sperber (1987), who hypothesized that these structures failed to develop as a result of a disturbance caused by an "interaction aplasia." In that study, organs or tissues failed to develop or were displaced in one twin because of conflicting migrational pathways or abnormal concentrations of morphogens in and around the two distinct organizing axes. In our case, we speculate that the embryonic mass that was formed as a consequence of an attempted twinning possibly had a hindgut anlage that responded to or "saw" the signal of only one organizing center or axis during subsequent development. This may have led to the autosite eventually ending up with a duplicated lower intestinal tract. It has also been suggested that the absence of a spinal axis in the parasite twin could be responsible for the absence of internal organs (Ursell and Wigger, 1983). Finally, we can further suggest that some vertebral structures and/or visceral organs might have been present in the early stages of postnatal life but either were not discernible grossly or subsequently disappeared with advanced age, because they were functionless.

Partial or "mild" twinning has been implicated as a cause that leads to the duplication of the lower intestinal and urogenital tracts where an obvious cotwin is lacking (Ravitch, 1953; Magbool et al., 1988). Other embryologic events, such as over development or duplication of the cloacal membrane (Hollowell et al., 1977), notochordal splitting (Bentley and Smith, 1960), and incomplete or delayed partitioning of the cloaca by the urogenital septum, are among the factors that have been implicated as induction factors for such defects (Gilsanz et al., 1982). In the present case, it is fairly clear that the effects of partial twinning were responsible for the observed duplications. Following the duplication of the primitive intestinal tube, portions of the midgut eventually gave rise to the jejunum and ileum, whereas the hindgut gave rise to the colon, rectum, and anal canal. The bladder and urethra were duplicated as well, because they arise from the proximal portion of the allantois and the cranial portion of the urogenital sinus, which are derivatives of the primitive hindgut (Noden and De Lahunta, 1985).

In female mammals, the paired paramesonephric (Mullerian) ducts give rise to the uterine tubes, uterus, cervix, and cranial portions of the vagina. The goat, like all other ruminants, has a bicornuate uterus in which the uterine horns join to form a uterine body that opens via the cervix into a single vagina. The separated paramesonephric derivatives in this case do not represent a true duplication when compared with the duplications of the lower urinary and intestinal tracts. The presence of duplex uterus, cervix, and vagina actually represented a total failure in fusion between the paired genital ducts at their caudal ends. In lower mammals (monotremes and most marsupials), this expression is normal, and the female ducts remain separated and open independently into the cloaca (Jarcho, 1946). In our case, the presence of a doubled urogenital sinus, to which the caudal end of the paramesonephric ducts normally attaches during early embryogenesis, mechanically prevented the fusion of the two paramesonephric ducts. In other words, the failure of these ducts to fuse is secondary to the primary splitting or duplication of the hindgut and its derivatives, allantois, bladder, and ureter. In the end, each duct developed independently and subsequently gave rise to a unicornous uterus and vagina. Unlike the internal genitalia, the

external genitalia are derivatives of the cloaca and the surrounding tissues; Because their anlage, the hindgut, had doubled, the external genitalia actually represented a true duplication.

The results of this study and those from human patients (Breen and Weinsburg, 1953: Beach et al., 1961) support the fact that, in higher mammals, both genital tracts can function normally, and lack of fusion or resultant separation does not render paramesonephric derivatives nonfunctional. Furthermore, in this case, both tracts were of normal size and morphology, whereas, in most human cases, it has been shown that there is a tendency for the left tract to be more developed than the right (Beach et al., 1961; Zamir et al., 1984). The PCT reported here had unique anatomical features that imply supernumerary rather than defective development and that were compatible with life. All of the affected organs attained normal development and function without evidence of defectiveness. The PCT presented in this study is another example of the anatomical variations whose developmentally sequelae are far from being fully explained.

## ACKNOWLEDGMENTS

We thank Dr. Alice F. Tarantal and Ms. Ruby A. Bell-Gam for their helpful comments during the preparation of this paper and Messrs. Christopher N. Kamwaro and Jackson M. Gachoka for the photography.

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