

Bifid phallus with complete duplication and a separate scrotum in a German shepherd dog: a case report

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ABSTRACT: Diphallia or penile duplication is a rare congenital abnormality thought to result from duplication of the cloacal membrane in the early developmental stages in the uterus. The present case concerned a 4-year old intact male German shepherd dog. The patient presented with a complaint of paraphimosis. Clinical, laboratory, radiological and ultrasonographic examinations were carried out. Semen samples collected prior to surgery were examined. Following amputation, the penis with paraphimosis was sent for histopathological investigation. Karyotype analysis of the patient was also performed. As well as the first report of this abnormality in Turkey, this case report represents the first report of this condition in a German shepherd dog.

Keywords: penile; abnormality; congenital; diphallia

Diphallia or penile duplication is a rare congenital abnormality and its incidence in humans is one in 5.5 million (Tirtayasa et al. 2013). Since it was first reported, there have been approximately 100 publications regarding this condition in human medicine (Tirtayasa et al. 2013; Galassi et al. 2016). However, it is also known that the condition has been observed in humans for thousands of years (Galassi et al. 2016). Diphallus has been observed and reported in cattle, dogs and donkeys (Bosu and Barker 1971; Johnston et al. 1989; Loynachan et al. 2006; Abu-Seida and Torad 2014). Only three cases have ever been reported in dogs (Potena et al. 1974; Johnston et al. 1989; Zucker et al. 1993). Diphallia may be caused by duplication of the cloacal membrane early in development, with subsequent formation of two urogenital tubercles, or may be part of a more extensive hindgut defect, which includes developmental anomalies in other systems (Hollowell et al. 1977).

Although penile duplication may be classified as orthotopic, ectopic or depending on the splitting characteristic of the penis, sagittal-frontal or

symmetrical-asymmetrical, it is generally classified into two main categories: true diphallia and bifid phallus. These categories are further divided into two subcategories: partial duplication and complete duplication. In true complete diphallia, there are two equal-sized penises with individual corpora cavernosa and corpus spongiosum; in true partial diphallia, on the other hand, one of the penises is either smaller or rudimentary. If there is only one corpus cavernosum in each penis, the condition is called bifid phallus. Bifid phallus is further subdivided into the following categories: complete, if there are two penises; and partial, if there are only two separate glandes (Gyftopoulos et al. 2002).

Case description

The present case concerned a 4-year-old intact male German shepherd dog. It was discovered that the two litter-mates of the patient did not have the same abnormality. Clinical examination revealed that the patient had two penises. The patient's own-

doi: 10.17221/16/2017-VETMED

ers informed the authors that they had adopted the dog at two months of age and had been aware of the two penises for four years; however, they had not considered the condition to be important. The reason for the visit to our clinic was that the penis, determined to be non-functional during clinical examination, was exposed from the prepuce and could not be retracted (paraphimosis). Clinical examination also revealed balanoposthitis. It was reported that the dog had never mated despite exhibiting interest.

In clinical examination, it was observed that the penis with paraphimosis ended under the skin lateral of the normal penis prepuce. The right testicle was in its normal anatomical position, while the left testicle was situated in separate pouches within the left inguinal canal (Figure 1A). Although glans penis, os penis and bulbus glandis were present in both penises, it was seen that the external urethra orifice had not developed in the penis with paraphimosis (Figure 1B). The functional penis had normal macroscopic anatomy and the external urethra orifice was present (Figure 1C). It could easily be catheterised using a male dog urethra catheter (Buster®, Kruuse). Complete blood count of the patient was performed, as well as determination of various serum biochemical parameters. Serum BUN, creatinine, ALT, AST and glucose values of the patient were normal. Mild leucocytosis ($18.1 \times 10^3 \mu\text{l}$, reference range 6–17) due to paraphimosis and balanoposthitis was present in the blood count. No radiographic abnormalities were encountered in the penile, pelvic or prostatic urethra of the patient during retrograde positive contrast urethrography. Radiographic examination revealed the os penis in the non-functional penis (Figure 2A). Ultrasound examination of the testicles and abdomen was carried out. The testicles

displayed a homogenous structure with regards to echogenicity and size, and no abnormalities were observed (Figures 2B and 2C). The kidneys were normal with regard to size, shape and density. There was no peri-renal fluid accumulation, hydronephrosis, cysts or tumours (Figures 2D and 2E).

Semen samples obtained from the patient via ejaculation were investigated. During examination, erection occurred only in the functional penis. Ejaculation via hand massage produced 0.8 ml semen. No sperm cells were seen in the microscopic examination of the semen. It was concluded that, even if the patient had mated, it would not be able to produce any offspring. The penis with paraphimosis was amputated and sent for histopathologic investigation. Castration was not performed.

Grossly, the glans penis was poorly developed and the external urethra orifice was not present in the glans penis. The penile urethra was not detected in randomly cut sagittal sections. For histopathology, tissue specimens were fixed in 10% neutral buffered formalin, treated with graded alcohols, embedded in paraffin, cut in 5- μm -thick sections, stained with haematoxylin and eosin (H&E) and then examined under a light microscope.

Microscopically, corpus spongiosum was present in the ventral area of the section; however, there was no urethral opening within this component (Figure 3A). Fibrinopurulent inflammation characterised by fibrin fibres, polymorphonuclear leukocyte infiltration and wide necrosis extending from the outer cutaneous mucosal layer deep into the corpus spongiosum were observed (Figure 3B). In the central regions of the sections, numerous hyperaemic venous vessels surrounded by a loose fibrous layer specific to the corpus cavernosa were observed (Figure 3C). There was abundant fibrin deposition

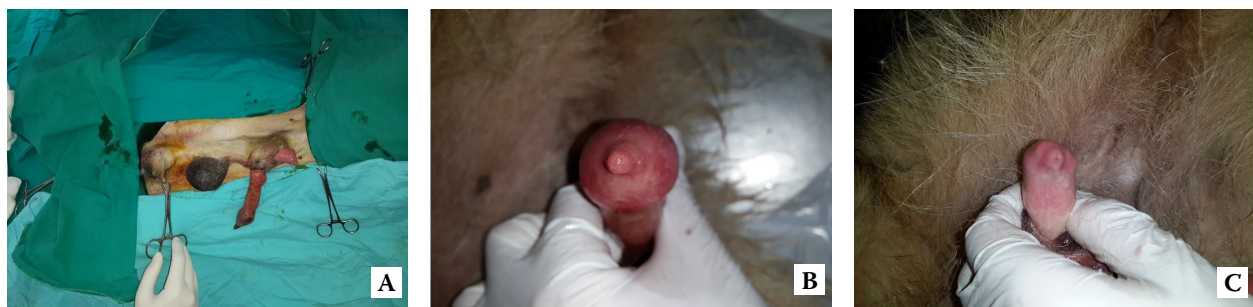


Figure 1. Appearance of the two penises and testicles within their separate scrotums (A). The glans penis of the non-functional penis and a perspective illustrating the lack of an external urethra orifice (B). The functional penis had normal macroscopic anatomy and the external urethra orifice was present (C)

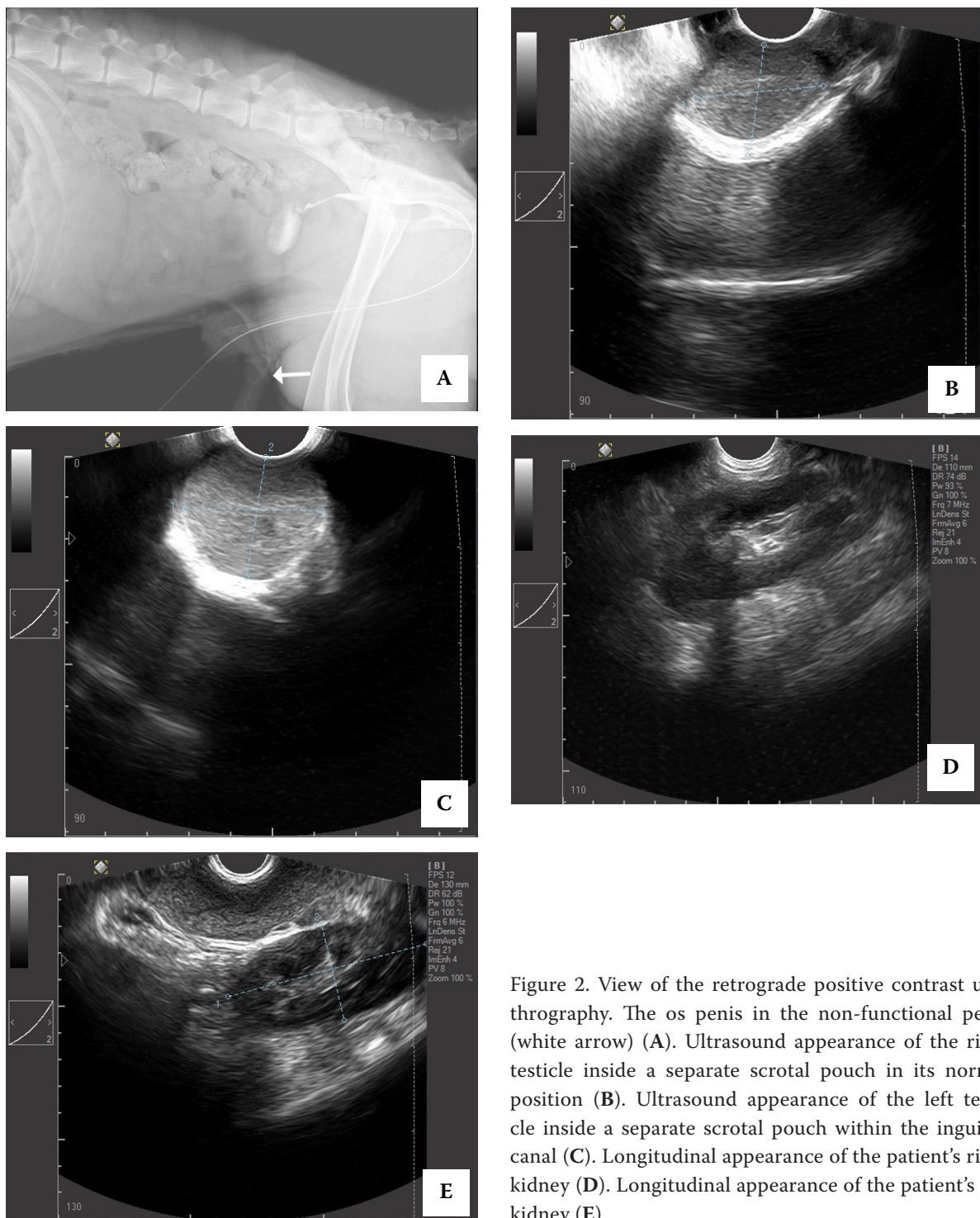


Figure 2. View of the retrograde positive contrast urethrography. The os penis in the non-functional penis (white arrow) (A). Ultrasound appearance of the right testicle inside a separate scrotal pouch in its normal position (B). Ultrasound appearance of the left testicle inside a separate scrotal pouch within the inguinal canal (C). Longitudinal appearance of the patient's right kidney (D). Longitudinal appearance of the patient's left kidney (E)

and numerous bacteria shaped like cocci in the necrotic areas (Figure 3D). Severe fibrinoid necrosis was apparent in both the walls of arterioles and the large venous vessels of corpus cavernosa, and thrombi were present within their lumina (Figure 3E). The stratified squamous epithelium layer was profoundly diminished. Although the corpus cavernosum con-

sists of two paired cylinders situated on the right and left side in a normal penile structure, there was only one cylinder situated centrally in the specimen. Based on histological evaluation, the amputated specimen was confirmed as a penile tissue with no urethra. Also, fibrinopurulent balanoposthitis was the predominant inflammatory reaction.

doi: 10.17221/16/2017-VETMED

Karyotyping analysis revealed that the patient had a 78, XY karyotype structure (Figures 4A and 4B). Therefore, this ruled out any relation to chromosomal abnormalities.

DISCUSSION AND CONCLUSIONS

Diphallia is a condition in which an individual is born with two penises. In humans, it is believed to be

a result of injury or chemical stress to the foetal mesoderm between 23 and 25 days of gestation. In the veterinary literature, only a handful of canine cases have been reported (Potena et al. 1974; Johnston et al. 1989; Zucker et al. 1993). It has not yet been confirmed whether the cause in canines is the same as in human cases. However, the fact that we did not encounter any chromosomal abnormalities in this patient may suggest that it could have developed due to reasons similar to those seen in humans.

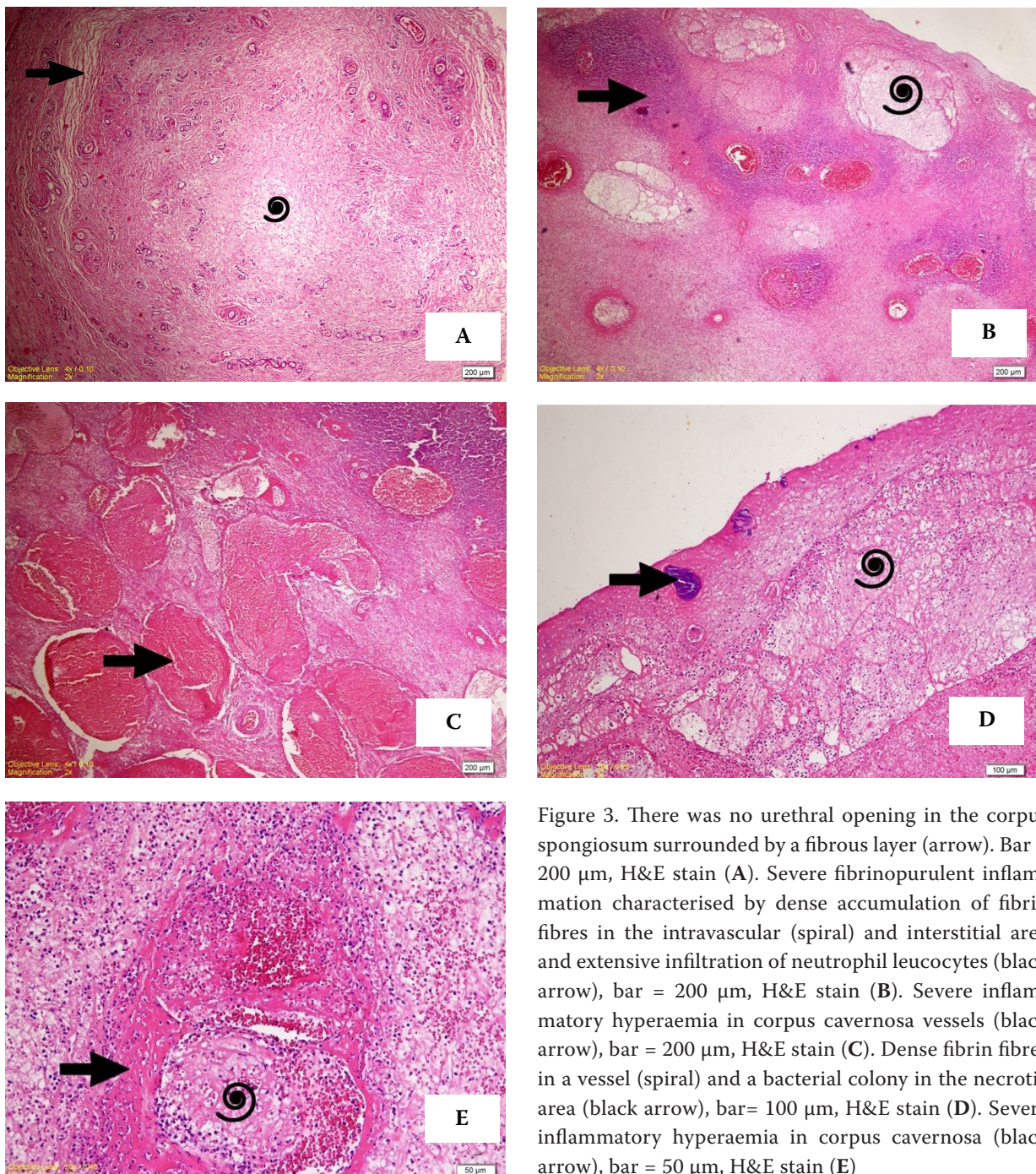


Figure 3. There was no urethral opening in the corpus spongiosum surrounded by a fibrous layer (arrow). Bar = 200 µm, H&E stain (A). Severe fibrinopurulent inflammation characterised by dense accumulation of fibrin fibres in the intravascular (spiral) and interstitial area and extensive infiltration of neutrophil leucocytes (black arrow), bar = 200 µm, H&E stain (B). Severe inflammatory hyperaemia in corpus cavernosa vessels (black arrow), bar = 200 µm, H&E stain (C). Dense fibrin fibres in a vessel (spiral) and a bacterial colony in the necrotic area (black arrow), bar = 100 µm, H&E stain (D). Severe inflammatory hyperaemia in corpus cavernosa (black arrow), bar = 50 µm, H&E stain (E)

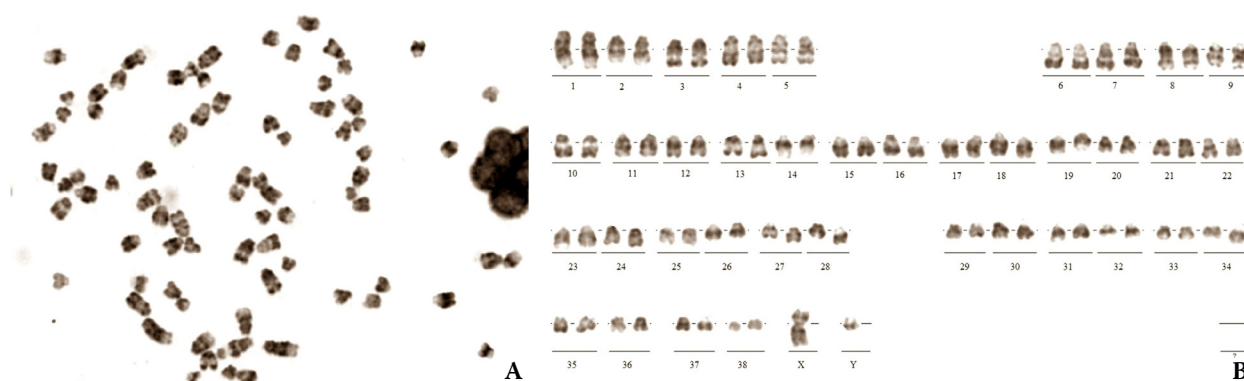


Figure 4. Metaphase chromosome spread (A). Karyotyping shows 78 chromosomes; chromosome constitution is 78, XY (B)

A telephone consultation with the patient's owner two years later revealed that the dog was in good health and that there was no haematuria or any symptoms related to the urinary system. The dog had not visited a veterinary practice in these subsequent two years except for routine vaccination, nor had it become unwell. The owner reported that the dog had not displayed any interest in mating during this time.

As well as the first diphallus case observed in Turkey, this report is also the first to describe the condition in a German shepherd dog. The only abnormality to be determined in the urogenital system, apart from the anatomical diphallus, was that the testicles were in separate positions and pouches. **Also, this is the first case in the veterinary literature in which karyotypic analysis was performed (Please, see Corrigendum, doi: 10.17221/16/2017-VETMED).**

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Received: January 30, 2017

Accepted after corrections: March 19, 2017