

Case Report

Incomplete Caudal Duplication With Correction of Pygomeia in a Dog



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**ABSTRACT**

Congenital malformations are structural defects that occur in all species during fetal development. Pygomeia is a type of polymelia in which the redundant legs are joined to the pelvis. This abnormality is observed in both human beings and animals.

A mixed female (Shih Tzu) puppy with a rigid mass resembling two pelvic hindlimbs was referred to a pet clinic in Mashhad City, Iran. In radiographic assessments, two extra hindlimbs stemmed from malformed pelvic bones and were shorter than normal limbs. Both extra acetabula were detected on the hemipelvis, with two extra coxofemoral joints perpendicular to the regular hip joints in the caudal direction. Also, two vulvas and two anuses were seen, but only one vulva and anus were functional.

The additional limbs were amputated by surgery. This case is the first report of a dog with incomplete caudal duplication and successful correction of pygomeia in Iran.

Keywords: Developmental anomaly, Hemipelvis, Shih Tzu, Twin, Surgery

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Introduction

A congenital abnormality is a defect or a set of defects in a growing fetus that could result in some structural or functional abnormalities. The exact etiology is unknown, but genetic or environmental factors or a combination of both are held accountable (DeSilva et al., 2016; Aliyu, 2021; Mozafari Makiabadi et al., 2022). Limb malformation is one of the most common congenital defects in animals and humans. These defects might be unilateral, multiple, or complex and can arise as a syndrome or in conjunction with other defects. Polymelia is the existence of additional limb(s), classified as cephalomelia (attached to the head), notomelia (attached to the vertebral column), thoracomelia (attached to the thorax), or pygromelia (attached to the pelvis) (Rahman et al., 2006; Shojaei et al., 2007). Pygromelia (complete limb duplication) is a rare abnormality characterized by the development of one or more extra limbs in the pelvic area (Bastiani-Griffet et al., 1990). Caudal duplication syndrome is characterized by total or partial duplication of the spine, spinal cord, and caudal anatomy, including the urogenital and gastro-

intestinal system, as well as neurological impairment to different degrees (Sur et al., 2013).

This paper describes a rare case of incomplete caudal duplication and pygromelia in a three-month-old female dog.

Case Presentation

A three-month-old female Shih Tzu dog, weighing 3.2 kg, with extra limbs in the pelvic region located under the tail, was admitted to a pet clinic (Figure 1). Clinical examination of posture and conformation of the dog reflected a healthy appearance, and it could walk and live normally with no clinical signs. However, it seemed uncomfortable to sit. The animal's nutritional and developmental statuses were good. In addition, heart rate, breathing frequency, and rectal temperature were 120 beats per minute, 22 breaths per minute, and 38°C, respectively. Her defecation was also normal in frequency, without evidence of distention or discomfort during the abdominal palpation. During the physical exam, the additional limbs were found to be static and smaller than the normal limbs. The flexion direction of joints was similar but



Figure 1. Two extra hindlimbs in a three-month-old female Shih Tzu dog



Figure 2. Ventrodorsal plain radiograph of pelvic region

Abbreviations: EAc: Extra acetabulum; EF: Extra femoral bone; ET: Extra tibia; EFi, extra fibula; EMT: Extra metatarsal bones.

Note: Extra hindlimbs are seen at the hemipelvis, as well as two extra coxofemoral joints, so ventral pelvic bones are not fused.

lower than the normal limbs. Although no neurological abnormalities were found in the four original limbs when pinching with forceps, her two extra rear limbs lacked innervation. Furthermore, there were two vulvas located right and left of the extra limb and an anal opening, as well as a tiny fold that resembled the dysplastic anus. Still, it did not have an opening (imperforate anus). On examination with a vaginoscope, it is notable that only one vulva was functioning, and patent and urine came out from it. The vagina of this genital tract was patent, and the existence of continuity between the vestibule and the cranial vagina was confirmed. Also, there was no connection between the duplicated genital structures.

Diagnostic Testing

In a radiographic evaluation, two extra hindlimbs originating from abnormal pelvic bones were observed, and the absence of fusion of the ventral pelvic bones was confirmed. The extra femur, tibia and fibula, tarsal bones, metatarsal bones, and digits were recognized on both sides by radiography. However, both extra acetabula located at the hemipelvis and two extra coxofemoral joints perpendicular to normal hip joints were observed in the caudal direction. The extra hindlimbs were shorter than normal limbs (Figure 2). A contrast radiographic via anal enema showed no sign of duplication in both descending colon and rectum, but a suspected small rectal/colon diverticulum was seen. It is hypothesized that this



Figure 3. Ventrodorsal contrast radiograph (barium enema)

Note: Duplication in the descending colon and rectum was excluded after the contrast study. But a suspected small rectal diverticulum was seen.

small diverticulum may be an incomplete gastrointestinal duplication (Figure 3).

In the current study, all blood counts and biochemical parameters were within the normal physiological range for dogs, and she was operated on to remove the additional legs. The prescribed prophylactic antibiotic and analgesia were cefazolin 500 mg (22 mg/kg) and meloxicam 2% (0.2 mg/kg), respectively. Acepromazine (0.08 mg/kg IV) was also used as a pre-anesthetic drug; ketamine 10% (6 mg/kg) and diazepam 10 mg/2 mL (0.2 mg/kg) were used for the induction. Moreover, isoflurane (MAC 2%) at oxygen 100% was used to maintain anesthesia. Finally, the pelvic region was prepared for surgery, and supportive fluid (20 mL/kg/h) was administered during the surgery.

An elliptical incision was initially made near the base of the additional limbs. After that, the subcutaneous tis-

sue was meticulously dissected, the muscles around the base limbs were carefully severed, the major vessels were ligated, and the tiny vessels were finally cauterized. We also found no nerve branches, and the additional limbs were observed to detach readily after reaching the basic bones, with no bone cuts. The remaining muscle tissues were closed with a simple continuous stitch, and the subcutaneous tissues were closed. Afterward, in an interrupted cross mattress, the skin was sutured using nylon (Figure 4), and the dog recovered smoothly. As the right vulva and dysplastic anus were nonfunctional and closed, so they were not annoying and remained intact. Since a small rectal/colon diverticulum was suspected, it remained intact until clinical signs appeared in the animal. Post-op treatment included the following items: tramadol (2 mg/kg, PO, every 8 hours, for 5 days) and cephalexin (25 mg/kg, PO, every 12 hours, for 7 days). The dissected removed limbs were smaller than her hindlimbs' bones, but all bones were intact. Femurs,



Figure 4. Extra limb removal

Note: True and dysplastic anus, left and right vulva are shown. Right nonfunctional vulva and dysplastic anus saved after surgery.

tibias and fibulas, metatarsals, and phalanges were completed with bone consistency (Figure 5). Bypassing two weeks after the surgery, the sutures were removed. The dog showed no symptoms of discomfort when sitting and defecating. There was no wound dehiscence with proper healing.

Assessments

Congenital malformations can be induced by genetic or environmental factors or both (Scholl & Thacker, 2021). Abnormalities in embryogenesis also cause these malformations. Furthermore, the birth defects might be related to the embryo's unequal distribution of germ cells or the abnormal duplication of those cells during embryo formation (Daneze & Brasil, 2018).

Pygomeia is a congenital musculoskeletal anomaly in which the extra limbs are joined to the pelvis via ru-

dimentary os coxae (Noh et al., 2003). Although pygomeia is generally caused by a caudal bifurcation of the body's long axis, it has also been observed in crossbred calves (Rahman et al., 2006; Mistry et al., 2010). These extra limbs with rigid joints and sparse muscles that lack innervation are always smaller than regular limbs (Talamillo et al., 2005). Since the cause of pygomeia is unknown, it could be classified as a dipygus separated into irregular splitting of a solitary embryo or two nosological problems because it is situated on the border of four other abnormalities such as sacrococcygeal tumors, lower limb duplications, double monsters, or isolated adjacent embryos with one that might display an insufficient development (Moulot et al., 2017). To remove extra limbs, however, a gradual and precise dissection of these redundant limbs under the supervision of competent preoperative and postoperative care would ensure a successful outcome. The case in this report suffered from pygomeia with no functional extra limbs. Consequent-

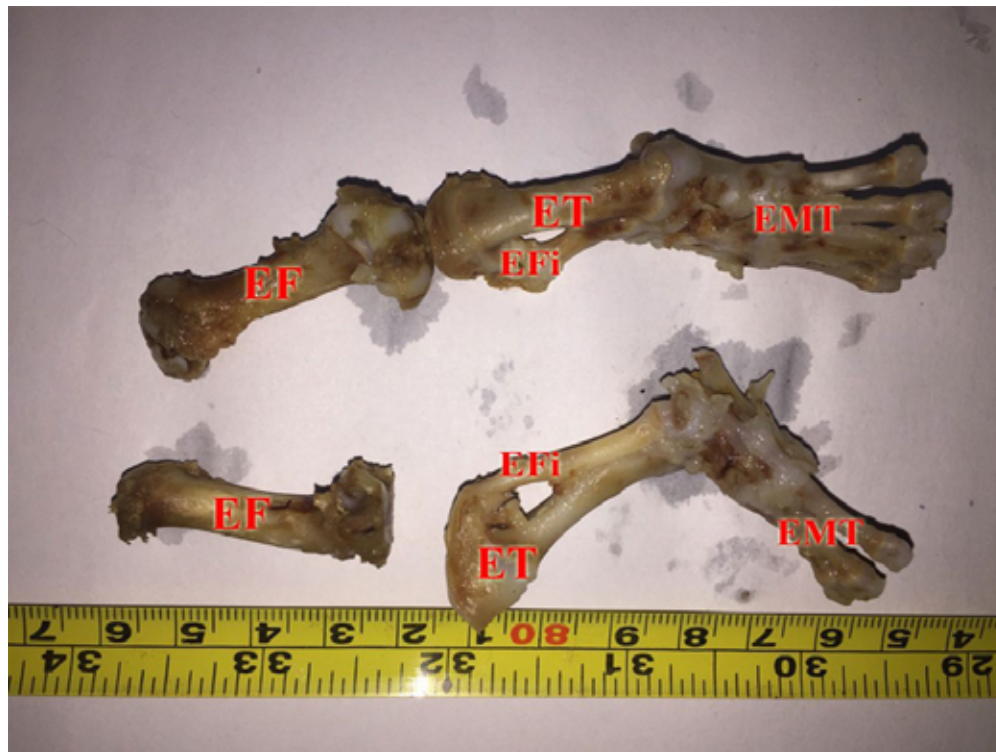


Figure 5. Dissected amputated two extra hindlimbs

Abbreviations: EF: Extra femoral bone; ET: Extra tibia; EFi: Extra fibula; EMT: Extra metatarsal bones.

ly, surgical intervention was advised because the extra limbs can interfere with adult growth and conformation and finally bring relief.

Conjoined twinning is formed by joining two more or less developed organisms, resulting in a duplication of the body axis (Schwalbe, 1907). This abnormality is defined by the existence of extra hind limbs linked to the pelvic area, as well as a duplication of pelvic organs. Twinning is complete if the zygote splits 8 to 13 days following fertilization (Harper et al., 1980). However, if the embryonic disk splits later, the division is typically partial, and the twins may be conjoined (Sarihan et al., 1980; Van den Brand et al., 1994). The presence of an accessory limb, considered a form of incomplete twinning, is referred to as dipygus or pygomeia. Pygomeia, pelvic organ, or spinal column duplications arise when they begin in the embryo's caudal region (Schwalbe, 1907).

The primary etiology and processes of caudal duplication and congenital limb abnormalities have remained unknown (Scholl & Thacker, 2021; Hirschberg et al., 2012). Beyond genetic changes, environmental variables such as *Lupinus* species consumption, viral infections, or exogenous hormone therapies are thought to induce congenital duplications (Murondoti & Busayi, 2001;

Kaufman, 2004). Another theory views over-aged oocyte ovulation as a potential cause (Witschi, 1970).

Incomplete forms of caudal duplications and pygomeia in humans (Moulot et al., 2017; Matthews et al., 1982; Bajpai et al., 2004; Alfadhel et al., 2009) and animals like cattle (Freick et al., 2014), cat (Seavers, 2009; Akbarian et al., 2020), dog (Mazzullo et al., 2007), pig (Ajadi & Olaniyi, 2018; Reiner et al., 2008), as well as in avian species like chicken (Hirschberg et al., 2012), pigeons (Corbera et al., 2012) and other species (Freick et al., 2014) have been published. In addition, the existence of ectopic and additional organs has been reported in some reports (Nazem et al., 2022).

A male crossbreed puppy was diagnosed with caudal duplication. In this case, an extra limb protrudes from the perineum, no tail, anal atresia, a double penis, and no scrotal sacs. An internal investigation revealed bowel and urine system abnormalities. The supernumerary limb's x-ray revealed the lack of the fibula and tibia, as well as the existence of three metatarsi and phalanges. The authors explored the pathogenic processes of this illness, which is seldom documented in veterinary practice, emphasizing the significance of embryonic duplications, typically linked with dystocia (Mazzullo et al., 2007).

Identifying and understanding the origin of congenital malformations remained a problem in veterinary practice, particularly given that most of these abnormalities occur infrequently and only in a few cases. However, it can become a research subject only in unusual circumstances, for example, when a similar problem occurs frequently within the same herd or geographic region. There were no documented incidents involving the progenitors. Besides, no medicines and hormone treatments were provided during gestation. No evidence was available to suggest that the duplication was environmentally or genetically induced or a combination of both. It is worth noting that this case was her mother's only pup.

Conclusion

In this case report, duplication of hindlimbs, vulva, and anus was evident. However, her extra vulva and anus did not function. Also, she had a suspected small rectal diverticulum that may not result in gastrointestinal duplication and could be considered an incomplete caudal duplication. This malformation probably occurred due to a defect in twin development during mother gestation. Since our case was young and healthy without the possibility of further intervention and re-anesthesia, we only successfully performed the amputation procedure without observing any sequelae or disability up to now. Although the incidence of this condition is extremely unusual, it should be examined using genetic testing, ultrasonography, CT-scan, magnetic resonance imaging, and other diagnostic imaging methods because this will provide us with more information about various diseases affecting the animals and allow us to make a more accurate diagnosis.

Ethical Considerations

Compliance with ethical guidelines

All ethical principles are considered in this article and the surgery was done with the consent of the animal owner.

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Authors' contributions

Conceptualization, methodology and validation: All authors; Investigation, resources and data curation: Shiva Amanollahi; Writing, visualization and supervision: Shiva

Amanollahi, Farzad Hayati and Ali Mirshahi; Project administration and funding acquisition: Farzad Hayati.

Conflict of interest

The authors declared no conflict of interest.

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گزارش موردی

دوتایی شدن خلفی ناقص همراه با اصلاح پیگوملیا در یک سگ

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چکیده

ناهنجاری‌های مادرزادی نقص‌های ساختاری هستند که در تمام گونه‌ها در طول رشد جنین رخ می‌دهد. پیگوملیا نوعی پلی ملیا است که در آن پاهای اضافی به لگن متصل می‌شوند. این اختلال در انسان و حیوانات مشاهده شده است. یک توله سگ ماده مختلط (شیتزو) با تودم‌های سفت که شبیه دو اندام عقبی در ناحیه لگن بود به کلینیک حیوانات خانگی در مشهد ارجاع شد. در ارزیابی‌های رادیوگرافیک، دو اندام خلفی اضافی از استخوان‌های بدشکل لگن منشأ گرفته و کوتاه‌تر از اندام‌های طبیعی بودند. هر دو استابولوم اضافی روی نیم لگن تشخیص داده شد و دو مفصل کوسوفمورال اضافی عمود بر مفاصل منظم هیپ در جهت خلفی یافت شدند. همچنین دو فرج و دو مقعد مشاهده شد که فقط یک فرج و مقعد کارکرد داشتند. در نهایت، اندام اضافی با عمل جراحی قطع شد. این اولین گزارش از یک سگ با دوتایی شدن خلفی ناقص و اصلاح موفق پیگوملیا در ایران است.

کلیدواژه‌ها: جراحی، دوقلویی، شیتزو، ناهنجاری رشد، نیم لگن



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