

## Incomplete Caudal Duplication with Correction of Pygomelia in a Dog

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### Abstract

Congenital malformations are structural defects that occur in all species during fetal  
15 development. Pygomelia is a type of polymelia in which the supernumerary legs are joined to the  
pelvis. It is observed in both human beings and animals. A mixed female (Shih Tzu) puppy with  
a rigid mass, which resembled two hindlimbs in the pelvic region was referred to a pet clinic in  
Mashhad. In survey radiographic assessments, two extra hindlimbs stemmed from malformed  
pelvic bones and were shorter than normal limbs. Both extra acetabulums were detected on the  
20 hemipelvis, and two extra coxofemoral joints were found 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. Finally, the additional limbs were amputated by surgery. This is the first report of a dog with incomplete caudal duplication and successful correction of Pygomelia in Iran.

**KEYWORDS:** Developmental anomaly; hemipelvis; shih Tzu; twin; surgery

## 25 *History*

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 causes are unknown, but, genetic or environmental factors or a combination of both are involved (DeSilva *et al.*, 2016; Aliyu, 2021; Mozaffari Makiabadi *et al.*, 2022). Limb malformation is one of the most common congenital  
30 defects in both animals and humans. They might be unilateral, multiple, or complex, and they can arise as a syndrome or in conjunction with other defects. Polymelia is described as the existence of additional limb(s), which may be classified as cephalomelia (attached to the head), notomelia (attached to the vertebral column), thoracomelia (attached to the thorax), or pygomelia (attached to the thorax) (attached to the pelvis) (Rahman *et al.*, 2006; Shojaei *et al.*, 2007).  
35 Pygomelia (complete limb duplication) is a rare abnormality defined 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 gastrointestinal system, as well as neurological impairment to different degrees. (Sur *et al.*, 2013).  
40 This paper described a rare case of incomplete caudal duplication and pygomelia in a three-month-old female dog.

## **Clinical Presentation**

A three-month-old female Shih Tzu dog, weighing 3.2 kg, with extra limbs in the pelvic region, which was 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 was able to walk normally with no clinical signs and with normal performance; however, it showed a sign of discomfort when sitting. The animal's nutritional and developmental statuses were at good levels. In addition, heart rate, breathing frequency, and rectal temperature were 120 beats per minute (bpm), 22 breaths per minute, and 38°C, respectively. As well, the defecation was normal in frequency and there was no 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 lower than the normal limbs. Although, when pinching with forceps, no neurological abnormalities were found in the four original limbs but two extra rear limbs lacked innervation. Furthermore, there were two vulvas that located right and left of the extra limb and an anal opening, as well as a tiny fold that resembled the dysplastic anus, but 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 survey by radiographic evaluations, 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 found on both sides

65 by radiography. However, both extra acetabulums located at the hemipelvis, were observed along with two extra coxofemoral joints perpendicular to normal hip joints, in the caudal direction. Of note, 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 small  
70 diverticulum may be an incomplete gastrointestinal duplication (Figure 3).

In the current study, all CBC 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,  
75 ketamine 10% (6 mg/kg), and diazepam 10 mg/2ml (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/hr) was administered during the surgery.

Firstly, an elliptical incision was made near the base of the additional limbs. Thereafter, the  
80 subcutaneous tissue 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 then closed with a simple continuous stitch and subsequently, the subcutaneous tissues were closed. Afterward, in  
85 an interrupted cross mattress, the skin was sutured using nylon (Figure 4), and the dog recovered smoothly. As regards, the right vulva and dysplastic anus were nonfunctional and closed, they

were not annoying, and remained intact. Since a small rectal/colon diverticulum was suspected, it also remained intact until clinical signs appear in the animal. Post-op treatment included the following items: tramadol (2 mg/kg, PO, every 8 hours, for 5 days) and cephalixin (25 mg/kg, PO, every 12 hours, for 7 days). When the dissected removed limbs were examined, although they were smaller compared to the bones of her hindlimbs, it was clear that all bones were intact. Femurs, tibias and fibulas, metatarsals, and phalanges were completed, which had bone consistency (Figure 5). Bypassing two weeks after the surgery, the sutures were removed. She showed no symptoms of discomfort with comfort when sitting and defecating. There was no wound dehiscence and healed properly.

### **Assessments**

*Congenital malformations can be induced by genetic or environmental factors or a mix of both. (Scholl and Thacker, 2021). These are also caused by abnormalities in embryogenesis. Furthermore, they might be related to the embryo's unequal distribution of germ cells or to the abnormal duplication of those cells during embryo formation. (Daneze and Brasil, 2018).*

*Pygamelia is a congenital musculoskeletal anomaly in which the extra limbs are joined to the pelvis via rudimentary os coxae. (Noh et al., 2003). It is generally caused by a caudal bifurcation of the body's long axis, while 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 (Rivera et al., 2005). Since the cause of pygamelia is unknown, it could be classified as a dypigies 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 supernumerary limbs under the supervision of competent preoperative and postoperative care would ensure a successful outcome. The dog described in this report suffered from pygomelia with no functional extra limbs. Consequently, surgical intervention was advised because the extra limbs can interfere with adult growth and conformation and finally bring relief.*

*Conjoined twinnings are formed by the joining of 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 between the days eighth and thirteenth 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 pygomelia. Pygomelia, or pelvic organ or spinal column duplications, arise when duplications begin in the embryo's caudal region. (Schwalbe, 1907).*

*The main etiology and processes of caudal duplication and congenital limb abnormalities remain unknown. (Scholl and 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 and Busayi, 2001; Kaufman, 2004). Another theory views over-aged oocyte ovulation as a potential cause (Witschi, 1970).*

130 *Incomplete forms of caudal duplications and pygomelia in humans (Moulot et al., 2017; Matthews et al., 1982; Bajpai et al., 2004; Alfadhel et al., 2009) and veterinary medicine like cattle (Freick et al., 2014), cat (Seavers, 2009; Akbarian et al., 2020), dog (Mazzullo et al., 2007), pig (Ajadi and Olaniyi, 2018; Reiner et al., 2008), as well as in avian species like chicken (Hirschberg et al., 2012), pigeons (Corbera et al., 2012) and in other species (Freick et al.,*  
135 *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, there is an extra limb protruding from the perineum, no tail, anal atresia, a double penis, and no scrotal sacs. An internal investigation revealed bowel and urine system abnormalities. The*  
140 *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 explore the pathogenic processes of this illness, which is seldom documented in veterinary practice, emphasizing the significance of embryonic duplications, which are typically linked with dystocia (Mazzullo et al., 2007).*

*Identifying and understanding the origin of congenital malformations remained a problem in*  
145 *veterinary practice, particularly given that most of these abnormalities occur infrequently and only in a few cases. However, only in unusual circumstances, such as when a problem frequently occurs within the same herd or geographic region, it can become a subject of research. 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*  
150 *duplication was environmentally or genetically induced, or a combination of both. It is worth noting that the pup was the only pup of her mother.*

## **Conclusion**

*In this case report, duplication of hindlimbs, vulva, and anus was evident. However, a vulva and anus did not function, also, a suspected small rectal diverticulum that maybe did not lead to gastrointestinal duplication can be considered as incomplete caudal duplication, which may be assumed that this malformation occurred due to a defect in twin development during mother gestation. Since our case was young and healthy and there was no possibility of further intervention and re-anesthesia, we only successfully performed the amputation procedure with no 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, MRI, 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.*

## **Acknowledgment**

*We thank the staff of the clinic for their cooperation in management of this case.*

## **Conflict of interest**

*The authors declare they have no conflicts of interest.*

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## 265 **Figure legends**

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

**Figure 2.** Ventrodorsal plain radiograph of pelvic region, extra hindlimbs are seen at the hemipelvis and two extra coxofemoral joints, so that ventral pelvic bones was not fused. EAc=

extra acetabulum, EF= extra femoral bone, ET= extra tibia, EFi= extra fibula, EMT= extra  
metatarsal bones.

**Figure 3.** Ventrodorsal contrast radiograph (barium enema). Duplication in descending colon and rectum was excluded after contrast study. But suspected small rectal diverticulum was seen.

**Figure 4.** Extra limb removed. True and dysplastic anus, Left and right vulva has showed. Right nonfunctional vulva and dysplastic anus saved after surgery.

**Figure 5.** Image shows dissected amputated two extra hindlimbs. EF= extra femoral bone, ET= extra tibia, EFi= extra fibula, EMT= extra metatarsal bones.

دوتایی شدن خلفی ناقص همراه با اصلاح پیگوملیا در یک سگ

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#### چکیده

ناهنجاری های مادرزادی نقص های ساختاری هستند که در تمام گونه ها در طول رشد جنین رخ می دهد. پیگوملیا نوعی پلی ملیا است که در آن پاهای اضافی به لگن متصل می شوند. این اختلال در انسان و حیوانات مشاهده شده است. یک توله سگ ماده مختلط (شیتزو) با توده ای سفت که شبیه دو اندام عقبی در ناحیه لگن بود به کلینیک حیوانات خانگی در مشهد ارجاع شد. در ارزیابی های رادیوگرافیک، دو اندام خلفی اضافی از استخوان های بدشکل لگن منشا گرفته و کوتاه تر از اندام های طبیعی بودند. هر

دو استابولوم اضافی روی نیم لگن تشخیص داده شد، و دو مفصل کوکسوفمورال 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.

کلمات کلیدی: جراحی؛ دوقلوئی؛ شیتزو؛ ناهنجاری رشد؛ نیم لگن





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



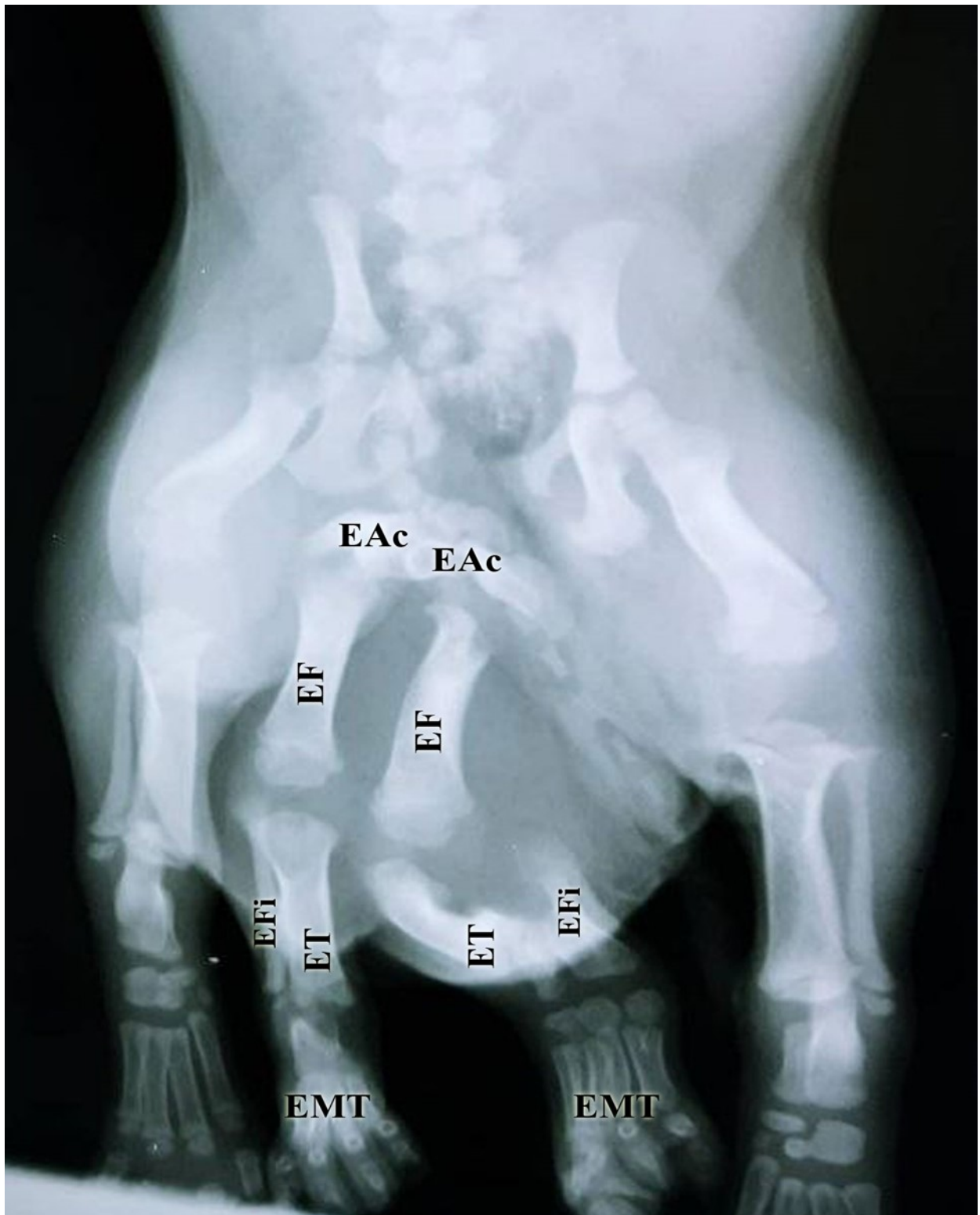


Figure 2. Ventrodorsal plain radiograph of pelvic region, extra hindlimbs are seen at the hemipelvis and two extra coxofemoral joints, so that ventral pelvic bones was not fused. EAc= extra acetabulum, EF= extra femoral bone, ET= extra tibia, EFi= extra fibula, EMT= extra metatarsal bones.



Figure 3. Ventrodorsal contrast radiograph (barium enema). Duplication in descending colon and rectum was excluded after contrast study. But suspected small rectal diverticulum was seen.

Uncorrected Proof



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