

## UNILATERAL LONGITUDINAL PREAXIAL RADIAL HEMIMELIA IN A PUPPY: A CASE REPORT

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### Abstract

Congenital deformities are structural or functional defects originating from errors during fetal development caused by various factors like use of chemotherapeutic and teratogenic drugs, malnutrition, transplacental viral infections, dietary mineral deficiencies, vaccines and they can affect a part of the body, entire body or parts of several systems of the body. Hemimelia is a congenital malformation of complete or partial absence of one or more bones. Treatment options depend on the extent of deformity and the reduction of the limb function. This report describes a case of unilateral longitudinal preaxial radial hemimelia, in a 20 days old female German Shepherd puppy and the starting of the treatment with Robert Jones splinted bandage to prevent further deformation and improve the limb function until bone maturity.

**Keywords:** *radial hemimelia; radiography; congenital malformations; dog*

### Introduction

Errors caused by various factors during fetal development may represent itself by structural or functional congenital malformations at birth (Noden and De Lahunta 1985). While these congenital malformations vary in different patterns, they can affect a single structure or function, parts of several systems or entire body (Dennis and Leipold 1979).

Congenital limb abnormalities can manifest in different patterns, ranging from the malformation of a single structure to partial or complete absence of the limbs (Lallo et al. 2001).

The insufficient precise nomenclature for limb malformations often complicates their description. Clinical case presented here was a type of hemimelia, a congenital abnormality with the complete or partial absence of one or more bones (Towle and Breur 2004). Longitudinal hemimelia is the congenital absence of one or more bones along the preaxial (medial) or postaxial (lateral) side of a limb (Bayne and Klug 1987; James and McCarroll 1999). Hemimelia can also classify as intercalary, which means all or part of the middle bones of a limb are absent, with the proximal and distal portions being present (Nelson and Blauvelt 2015).

Also, hemimelia can be classified as transverse, with a complete absence of the distal portion of the limb, and paraxial characterized by aplasia of either the radius or ulna or tibia and fibula (Nelson and Blauvelt 2015).

The aim of this study was to describe the clinical and radiographic findings of a rare case of a unilateral forelimb malformation observed in a puppy, and preserve the limb while improving its function until bone maturity.

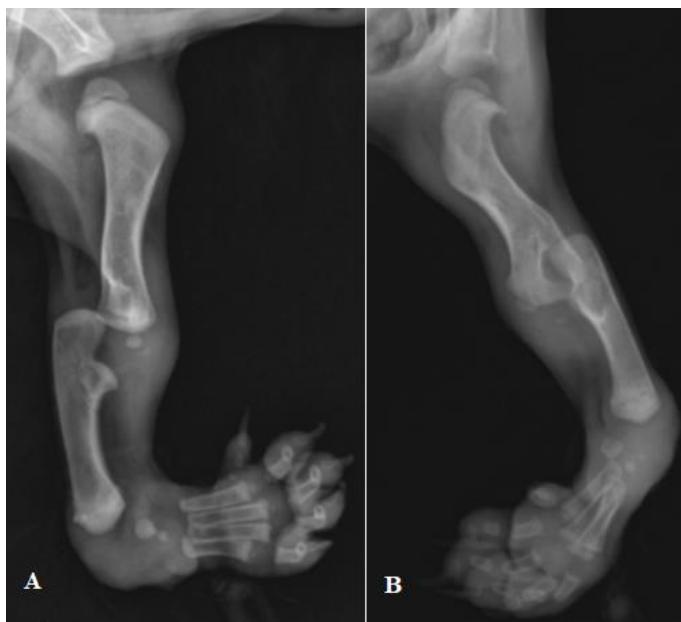
### Case Presentation

20 days old female German Shepherd puppy presented to the Department of Surgery, Faculty of Veterinary Medicine, Ankara University, for an orthopedic examination of a deformity of the left forelimb. The limb defect had been present since birth and a non-functional left forelimb, reduced in length with varus deviation on the carpal joint was observed on examination (Figure 1). There was no sign of pain, no crepitus or sign of fractures on physical examination.



**Figure 1.** Deficient left forelimb characterized by the reduced in length and varus deviation on the carpal joint.

Radiographs showed agenesis of the radius, poor congruency of the humeroulnar and carpal joints (Figure 2). The resulting diagnosis was unilateral longitudinal preaxial radial hemimelia.



**Figure 2.** Mediolateral (A) and anteroposterior (B) radiographs of the left forelimb. Note the congruency of the humeroulnar and carpal joints.

Treatment commenced with Robert Jones splinted bandage with one splint with a thickness of 3 mm on the posterior surface of the limb as support. The splint length was the same as the length of the forearm and the width was 6 mm (Figure 3).

The aim was to reduce the deformity of the limb to a level close to normal limb alignment with a splinted bandage until bone maturity. Initially, the dog was uncomfortable with the bandage and took a while it got used to the altered position. Each time the bandage did not fit adequately, it was restored, usually once a week. The only complication developed by the bandage was a small wound caused by the nail of the first phalanx.



**Figure 3.** Dog with Robert Jones splinted bandage

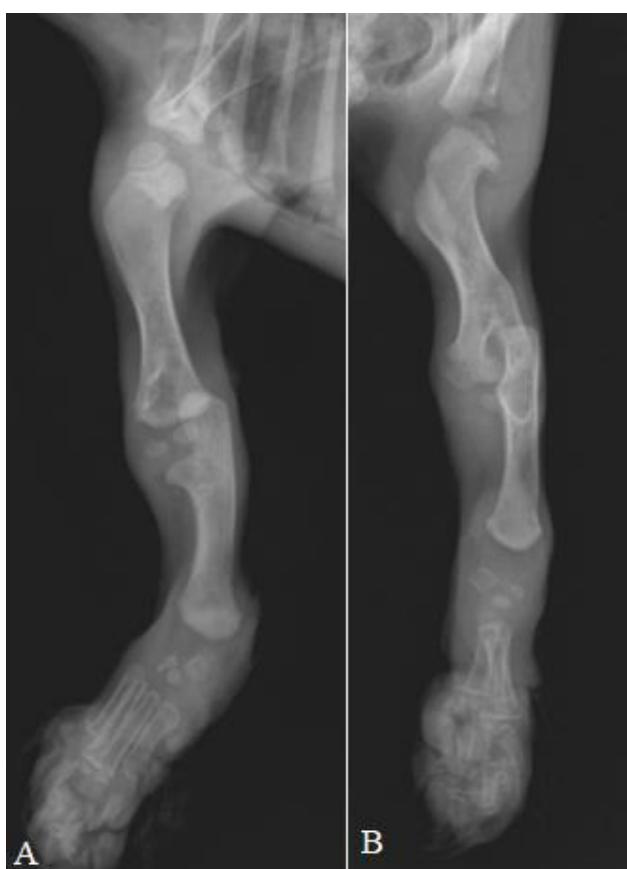


**Figure 4.** Two weeks after starting the treatment. Note that the angulation of the carpal joint getting closer to the acceptable degree.

After three weeks of treatment, the forearm was very close to normal and despite the reduced size of the limb, and congruency of the humeroulnar joint, the dog was trying to use it when standing and walking(Figures 5 and 6).



**Figure 5.** After three weeks of bandage application, the angulation of the carpal joint was in an acceptable range and despite the reduced length of the limb, the dog was trying to use it.



**Figure 6.** Mediolateral (A) and anteroposterior (B) radiographs of the left forelimb after three weeks. Note the slight curvature of the ulna and retaining the congruency of the humeroulnar joint.

### Discussions and Conclusions

Various patterns of congenital lower extremity deformities have been reported in dogs and cats: adactyly (Barrand and Cornillie 2008), aphalangia (Di Dona and Della Valle 2016), brachymelia (Cornillie et al. 2004), ectromelia (De Lima 1915; Macri et al. 2009), hemimelia (Pedersen 1968; Lewis and Van Sickle 1970; Alonso et al. 1982; Schultz and Watson 1995; Lallo et al. 2001; Rahal et al. 2005; Alam et al. 2006), radial agenesis (Lewis and Van Sickle; 1970 Swalley and Swalley 1978; Richardson 1979; Betts 1981; Winterbotham et al. 1985;

O'Brien et al. 2002; Rahal et al. 2005; Hildreth and Johnson 2007; McKee and Reynolds 2007), syndactyly (Hays 1917; Towle and Blevins 2007).

Possible causes of hemimelia include the administration of chemotherapeutics and teratogenic drugs, malnutrition (riboflavin deficiency), transplacental viral infections, dietary mineral deficiency (zinc, manganese, copper, etc.) and vaccines (Johnson 1965; Karnofsky 1965; Warkany 1965) during pregnancy.

Alonso et al. (1982) and Hoskins (1995) suggested that hemimelia may be hereditary due to an autosomal recessive inheritance. Mutation of En-1, FGF-2, Wnt7a, Shh, and Lmx-1 genes have been linked to the development of hemimelia in human, chick and mouse embryos (Chiang et al. 2001; Towle and Breur 2004; Woods et al. 2006). Also, hemimelia may result from a lack of apical ectodermal ridge - mesodermal interaction during limb outgrowth (Rantanen and Hegreberg 1982; Ogden and Grogan 1987; Towle and Breur 2004).

Between the 3rd and 4th weeks of pregnancy is the critical period for limb development in the canine embryo, when tissues are more susceptible to external influences (Noden and De Lahunta 1985).

In this case, our goal was to preserve the limb and improve its function until the bone maturity. Considering the weight of an adult German Shepherd, surgical procedures will be evaluated.

### References

Ahalt BA, Bilbrey SA (1997): What is your diagnosis? Radial agenesis with secondary contractual deformity of the right forelimb. *Journal of Small Animal Practice* 38, 539– 571.

Alam MR, Heo SY, Lee HB, Kim JH, Park YJ, Lee KC, Choi IH, Kim NS (2006): Preaxial longitudinal intercalary radial hemimelia in a dog: a case report. *Veterinarni Medicina* 51, 118–123.

Alonso RA, Hernandez A, Diaz P, Cantu JM (1982): An autosomal recessive form of hemimelia in dogs. *Veterinary Record* 110, 128–129.

Barrand KR, Cornillie PK (2008): Bilateral hindlimb adactyly in an adult cat. *Journal of Small Animal Practice* 49, 252–253.

Betts CW (1981): Agenesis of the radius. In: Bojrab MJ, Wingfield WE, Olds RB (eds): *Pathophysiology in Small Animal Surgery*. Lea and Febiger, Philadelphia. 650–653.

Bayne, L. G., Klug, M. S. (1987). Long-term review of the surgical treatment of radial deficiencies. *The Journal of Hand Surgery*, 12(2), 169–179.

Bezhentseva, A., Singh, H., & Boudrieau, R. (2018). Bilateral Radial Agenesis in a Cat Treated with Bilateral Ulnocarpal Arthrodesis. *Veterinary and Comparative Orthopaedics and Traumatology*.

Chiang C, Litingtung Y, Harris MP, Simandl BK, Li Y, Beachy PA, Fallon JF (2001): Manifestation of the limb prepattern: limb development in the absence of sonic hedgehog function. *Developmental Biology* 236, 421–435.

Cornillie P, Van Lancker S, Simoens P (2004): Two cases of brachymelia in cats. *Anatomia Histologia Embryologia* 33, 115–118.

De Lima JA (1915): On the skeleton of an ectromelic goat. *Journal of Anatomy and Physiology* 49, 378–385.

Dennis SM, Leipold HW (1979): Ovine congenital defects. *Veterinary Bulletin* 49, 233–239.

Di Dona, F., Della Valle, G., Meomartino, L., Lamagna, F., & Fatone, G. (2016). Congenital deformity of the distal extremities in three dogs. *Open Veterinary Journal*, 6(3), 228.

Gemmill TJ, Clarke SP, Carmichael S (2004): Carpal agenesis in a domestic short haired cat. *Veterinary and Comparative Orthopaedics and Traumatology* 17, 163–166.

Hays, G. P. (1917) A case of syndactylyous cat. *Journal of Morphology* 30, 65-82

Hildreth BE, Johnson KA (2007): Ulnocarpal arthrodesis for the treatment of radial agenesis in a dog. *Veterinary Comparative Orthopaedics and Traumatology* 20, 231–235.

Hoskins J (1995): Congenital defects of cats. *Compendium on Continuing Education for the Practicing Veterinarian* 17, 385–405.

James, M. A., McCarroll, H. R., & Manske, P. R. (1999). The spectrum of radial longitudinal deficiency: A modified classification. *The Journal of Hand Surgery*, 24(6), 1145–1155.

Johnson EM (1965): Nutritional factors in mammalian teratology. In: Wilson JG, Warkany J (eds): *Teratology, Principles and Techniques*. The University of Chicago Press, Chicago. 113–130.

Johnson KA, Watson ADJ, Page RL (1995): Skeletal diseases. In: Ettinger SJ, Feldman EC (eds): *Textbook of Veterinary Internal Medicine*. 4th edn. Saunders W.B., Philadelphia. 2077–2102.

Karnofsky DA (1965): Mechanism of action of certain growth inhibiting drugs. In: Wilson JG, Warkany J (eds): *Teratology, Principles and Techniques*. The University of Chicago Press, Chicago. 185–214.

Lallo MA, Bondan EF, Xavier JG, Fernandes TP, Kolber M, Zanco NA (2001): Bilateral anterior hemimelia in a dog: a case report. In: 26th World Small Animal Veterinary Association (WSAVA) World Congress. Vancouver, British Columbia, Canada. August 8.–11., 2001.

Lewis, R. E. & Van Sickle, D. C. (1970) Congenital hemimelia (agenesis) of the radius in a dog and cat. *Journal of the American Veterinary Medical Association* 156, 1892–1897

Macri F, De Majo M, Rapisarda G, Mazzullo G (2009): Two cases of feline ectromelia: autopodium lia associated with humero-ulnar synostosis and zeugopodium ectromelia. *Journal of Feline and Medicine Surgery* 11, 731–734.

Macri F, Marino F, Rapisarda G, Lanteri G, Mazzullo G (2011): A case of unilateral pelvic limb adactyly in a puppy dog. *Anatomia Histologia Embryologia* 40, 104–106.

Macri F, Ciotola F, Rapisarda G, Lanteri G, Albarella S, Aiudi G, Liotta L, Marino F (2013a): A rare case of simple syndactyly in a puppy. *Journal Small Animal Practice* 55, 170–173.

Macri F, Lanteri G, Rapisarda G, Marino F (2013b): Unilateral forelimb partial aphalangia in a kitten. *Journal of Feline and Medicine Surgery* 14, 272–275.

Macri F, Rapisarda G, Lanteri G, Di Pietro S, Auriemma E, Marino F (2014): Congenital absence of humerus with preaxial terminal longitudinal hemimelia and hypoplasia of the scapula in a dog: a case report. *Veterinarni Medicina* 59, 506–510.

Macri, F., Di Pietro, S., Palumbo Piccionello, A., Rapisarda, G., Lanteri, G., Angileri, V., & Marino, F. (2017). A rare case of partial paraxial radial hemimelia in a puppy. *Veterinární Medicína*, 62(No. 12), 681–684.

McKee WM, Reynolds J (2007): Ulnocarpal arthrodesis and limb lengthening for the management of radial agenesis in a dog. *Journal of Small Animal Practice* 48, 591–595.

Nelson, F. R. T., & Blauvelt, C. T. (2015). *Musculoskeletal Diseases and Related Terms. A Manual of Orthopaedic Terminology*, 43–104.

Noden DM, de Lahunta A (eds) (1985): *The Embryology of Domestic Animals, Development Mechanisms and Malformations*. Williams Wilkins, Baltimore. 367 pp.

O'Brien CR, Malik R, Nicoll RG (2002): Bilateral forelimb deformity and abnormal gait in a young Devon Rex. *Journal of Feline Medicine and Surgery* 4, 112–113.

Ogden JA, Grogan DP (1987): Prenatal skeletal development and growth of the musculoskeletal system. In: Albright JA, Brand RA (eds): *The Scientific Basis of Orthopedics*. Appleton and Lange, Norwalk. 47–89.

Palmer N (ed.) (1993): Pathology of Domestic Animals. 4th edn. Academic Press, San Diego. 45–47.

Pedersen NC (1968): Surgical correction of a congenital defect of the radius and ulna of a dog. *Journal of the American Veterinary Medical Association* 153, 1328–1331.

Pisoni L, Cinti F, Del Magno S, Joechler M (2012): Bilateral radial hemimelia and multiple malformations in a kitten. *Journal of Feline and Medicine Surgery* 14, 598–602.

Rahal SC, Volpi RS, Ciani RB, Vulcano LC (2005): Use of the Ilizarov method of distraction osteogenesis for the treatment of radial hemimelia in a dog. *Journal of the American Veterinary Medical Association* 226, 65–68.

Rantanen NW, Hegreberg GA (1982): Naturally occurring radial aplasia in mink. *Veterinary Radiology* 23, 27–29.

Richardson DC (1979): Radial agenesis. *Journal of Veterinary Orthopaedics* 1, 39–42.

Riddle Jr WE, Leighton RL (1970): Osteochondromatosis in a cat. *Journal of American Veterinary Medical Association* 156, 1428–1430.

Schultz VA, Watson AG (1995): Lumbosacral transitional vertebra and thoracic limb malformations in a Chihuahua puppy. *Journal of American Animal Hospital Association* 31, 101–106.

Swalley J, Swalley M (1978): Agenesis of the radius in a kitten. *Feline Practice* 8, 25–26.

Towle H.A., Breur G.J. (2004): Dysostosis of the canine and feline appendicular skeleton. *Journal of the American Veterinary Medical Association* 225, 1685–1692.

Towle, H. A., Blevins, W. E., Tuer, L. R. & Breur, G. J. (2007) Syndactyly in a litter of kittens. *Journal of Small Animal Practice* 48, 292-296

Warkany J (1965): Development of experimental mammalian teratology. In: Wilson JG, Warkany J (eds): *Teratology, Principles and Techniques*. The University of Chicago Press, Chicago. 1–12.

Winterbotham EJ, Johnson KA, Francis DJ (1985): Radial agenesis in a cat. *Journal of Small Animal Practice* 26, 393–398.

Woods CG, Stricker S, Seemann P, Stern R, Cox J, Sheridan E, Roberts E, Springell K, Scott S, Karbani G, Sharif SM, Toomes C, Bond J, Kumar D, Al-Gazali L, Mundlos S (2006): Mutations in WNT7A cause a range of limb malformations including Fuhrmann syndrome and Al-Awadi/Raas-Rothschild/Schinzel Phocomelia syndrome. *American Journal of Human Genetics* 79, 402–408.