Preaxial longitudinal intercalary radial hemimelia in a dog: a case report

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ABSTRACT: A 9-month-old 10 kg intact male crossbred dog was admitted to The Animal Medical Centre, Chonbuk National University with congenitally malformed right forelimb. Physical exanimation of the limb showed varus deviation of the forepaw and flexion contracture of the interphalangeal joints. The survey radiographs revealed complete absence of the radius, poor congruency of the humeroulnar joint and 90° varus deviation of the ulnocarpal joint. A preaxial longitudinal intercalary radial hemimelia was diagnosed and the dog underwent 3 surgeries for correction of the deformity. During the first surgery, olecranon osteotomy and arthrodesis of the elbow were performed. Three months later, a second surgery was done to perform carpal panarthrodesis using bone plate and screws which was unsuccessful. Two weeks after, with a third surgery carpal arthrodesis was performed using acrylic frame, the distal portion of limb was straightened and the residual deviation of the forepaw was corrected. The surgical corrections resulted in satisfactory use of the limb and a better quality of life.

Keywords: radius; hemimelia; arthrodesis; dog

Malformations of the extremities or parts of them are varied in their manifestations, ranging from absence of a single structure to partial or complete absence of the limbs (Lallo et al., 2001). Radial hemimelia is a congenital abnormality characterized by the partial or complete absence of the radius (Corbera et al., 2002; Towle and Breur, 2004). If all or part of the middle bones of a limb are absent, with the proximal and distal portions being present, the hemimelia is called intercalary. The longitudinal hemimelia indicates the absence of one or more bones along the preaxial (medial) or postaxial (lateral) side of the limb (Mo and Manske, 2004; Towle and Breur, 2004). Preaxial longitudinal intercalary radial hemimelia is the most common type of hemimelia in dogs and cats. This condition is usually unilateral, but bilateral absence may occur. The condition is usually noticed soon after birth (Towle and Breur, 2004). In human preaxial radial hemimelia is considered to be sporadic or very rare; the reported incidence of the deformity in human is approximately 1 per 1 million of live births (Lenz, 1980).

Arthrodesis is a moderately disabling procedure that should be considered as an alternative to amputation. The most common indications for elbow arthrodesis are degenerative joint disease secondary to ununited anconeal process, fragmented coronoid process, osteochondrosis dissecans, chronic luxation or subluxation (traumatic and congenital), irreparable articular fracture, nonunion and malunions and asynchronous radius-ulna growth (Newton and Nunamaker, 1984; Lee at al., 2005). Arthrodesis is performed more often in the carpus than any other joints and the limb function is not adversely affected (Johnson, 1981). A number of fixation methods for arthrodesis have been described such as the bone plates, screws, cross pins, tension band wiring, ring fixators, and external skeletal fixation used alone or in combination (Newton and Nunamaker, 1984; Lee et al., 2005). The use of acrylic connecting bar external fixators has become widespread in veterinary orthopedics. This is the result of low cost of the materials, the simplicity of the technique, and its adaptability to a variety of pin diameters. An acrylic connecting

bar can easily be contoured to the shape of the body and allows the surgeon to place transcortical pins in different planes (Shahar, 2000). Autogenous cancellous bone grafts are used in an arthrodesis to stimulate osseous union and to shorten healing time, so that joint fusion occurs before implant loosening or failure (Johnson, 1981). The purpose of this study is to evaluate the surgical technique, complications and final functional outcomes of surgical management of hemimelia with elbow arthrodesis and carpal panarthrodesis using acrylic frame external fixator in a dog.

CASE PRESENTATION

A 9-month-old 10 kg intact male crossbred dog was admitted to The Animal Medical Centre, Chonbuk National University for the diagnosis and treatment of a deformity of the right forelimb that had been present since birth. In physical exanimation, a nonfunctional right forelimb, varus deviation of the forepaw and flexion contracture of the interphalangeal joints were observed (Figure 1). There was decreased range of motion of the right elbow joint and moderate disuse muscle atrophy with no pain during palpation. No other coexistent clinical abnormality was found on physical examination. The left limb was clinically normal. The survey radiographs revealed a poor congruency of the humeroulnar joint, complete absence of the radius, 90° varus deviation of the ulnocarpal joint, misshapen carpal bones, and flexion deformity of the interphalangeal joints (Figure 2). Serum biochemistry and complete blood count analyses were within reference values. The diagnosis was made as preaxial longitudinal intercalary radial hemimelia.

After premedicated with atropine sulphate (Atropine Sulfate Inj®, Dai Han Pharm. Co. Ltd., Korea) 0.05 mg/kg, s.c., the anaesthesia was induced using thiopentone sodium (Thionyl Inj®, Dai Han Pharm. Co. Ltd., Korea) 25 mg/kg, i.v. and maintained with enflurane and oxygen delivered through a cuffed endotracheal tube. Supportive fluid therapy was maintained throughout the procedure and cephalexin (Methilexin Inj®, Union Korea Pharm. Co. Ltd., Korea) 25 mg/kg, i.v. was administered at the time of induction.

Figure 1. Photograph of a 9-month-old male dog showing the right radial hemimelia with the varus deviation of the forepaw and flexion contracture of the interphalangeal joints

Figure 2. Preoperative radiograph of the right forelimb revealed a poor congruency of the humeroulnar joint, complete absence of the radius, 90° varus deviation of the ulnocarpal joint, misshapen carpal bones, and flexion deformity of the interphalangeal joints

The patient was positioned on dorsal recumbency and draped. A lateral incision was made and blunt dissection of the muscles and tissues were performed to expose the elbow joint. Osteotomy of the olecranon process was performed. The anconeus muscle and lateral joint capsule were incised to expose the humeroulnar joint. Articular cartilage was removed from the joint surface, the ulna was lightly rotated laterally and temporary fixation of the arthrodesis was maintained with a Kirschner wire. A bone plate was contoured and applied to the caudal surfaces of the humerus and ulna. Autogenous corticocancellous bones were collected from the osteotomized olecranon process and harvested at the arthrodesis site. The wound was closed in a usual manner. The optimal angle of the joint following arthrodesis appeared to be 130°. The limb was immobilized in a spica splint for four weeks with exercise restriction until radiographic fusion was evident.

Twelve weeks after, with the same anaesthetic protocol, a second surgery was done to perform carpal panarthrodesis. Surgical exposure was accomplished using a cranial incision from mid ulna, across the carpus to midmetacarpus. All three joints of the carpus were opened transversely to expose the articular surfaces. The articular cartilage was debrided from all surfaces to the depth of bleeding subchondral bone. A bone plate was contoured and was applied first to the cranial surface of the ulna and positioned so that one screw can be placed into the radial carpal bone. After radial attachment, the plate was positioned over and attached to the third metacarpal bone. The central holes of the plate, which lie over the carpus, were the last to be filled with screws.

Two weeks later it was complicated with loosening of screws and wound formation. With a third surgery the bone plate and bone screws were removed and an acrylic frame was used to perform carpal panarthrodesis, the distal portion of limb was straightened and the residual deviation of the forepaw was corrected. The ulna and metacarpal bones were exposed and two fixator pins were inserted on the ulnar shaft and another two across the metacarpal bones in a mediolateral direction and the pins were left long enough to attach into a temporary connecting bars and a temporary stabilization was achieved. The arthrodesis sites were filled with autogenous cancellous bone. The wound was closed in a routine manner. On either side, corrugated plastic tubing that will mold the connecting bars was pushed over the fixation pins and pushed towards the clump. The dependent ends of the tubing were plugged with cotton wadding. Methyl methacrylate powder and liquid monomer (2:1) were mixed with a tongue depressor in a disposable paper cup. The liquid Methyl methacrylate mixture was taken into a catheter tip 60-ml syringe and was injected into the flexible tubing, taking great care to ensure regular filling and to avoid trapping air bubbles (Piermattei and Flow, 1997; Kraus et al., 2003). After hardening of the acrylic bars, the temporary connecting bars were removed (Figure 3).

The postoperative treatment was given with cephalexin (Methilexin Inj®, Union Korea Pharm. Co. Ltd., Korea) 25 mg/kg, i.v, every 8 hrs, for 5 days and Dexamethasone (Dexamethasone Inj®, Daewon Pharm. Co. Ltd., Korea) 0.2 mg/kg, i.v., every 6 hrs for 3 days after each surgery.

After the surgical corrections the condition of the limb was monitored by physical examination and radiography. Radiograph of the elbow taken 6 weeks after the first surgical correction revealed a complete healing and fusion of the joint. One month after elbow arthrodesis the dog tried to bear weight on the affected limb but could not because of the residual deviation of the paw. After carpal arthrodesis with bone plate and screws, the joints become painful and there was exudation. However, carpal panarthrodesis with the application of acrylic frame external fixator resulted in improvement of the condition of the limb and a complete arthrodesis was observed after 8 weeks.

Figure 3. Carpal panarthrodesis using acrylic frame external fixator

Figure 4. Postoperative radiograph of the right forelimb taken 10 weeks after the final surgery revealed a complete elbow and carpal arthrodesis

After 10 weeks of the carpal arthrodesis, the acrylic frame was removed and a functional arthrodesis was observed and no instability was found on physical examination (Figure 4). During the final evaluation 10 months after the final surgery, the dog had an improvement of its gait and could partially bear weight on the affected limb when standing (Figure 5). However, the limb was a little shorter than the contralateral limb.

DISCUSSION

Congenital absence of radius is not common in dog and cats (Winterbrotham et al., 1985; Jonson, 1995). Congenital deficiencies of the forelimb include segmental deficiencies of the radius and ulna (Lewis and Van Sickle, 1970). The affected limb(s) are shortened with a varus deformity and are not functional or gaiting (Jonson, 1995). In our patient, the clinical signs were also similar with the previous reports of hemimelia in dogs (Lallo et al., 2001; Rahal et al., 2005), cats (Lewis and Van

Figure 5. Photograph taken 10 months after the final surgery revealed an improved appearance of the right forelimb

Sickle, 1970), goats (Corbera et al., 2002), minks (Rantanen and Hegreberg, 1982), and humans (Mo and Manske, 2004). The etiology of radial hemimelia is considered to be genetical, environmental or a combination of both factors. The genetical defects that cause the limb deficiencies are associated with autosomal dominant inheritance; however, other genetical causes include an autosomal recessive inheritance and chromosomal aberrations (Lenz, 1980; Alonso et al., 1982). Also different teratogenic agents have been related to congenital defects (Jonson, 1995; Ogino, 2004). In dogs and cats, limb formation occurs from day 23 of gestation to approximately day 35 (Towle and Breur, 2004). Congenital malformations of the forelimb occur more frequently than those of the hindlimb, as the forelimb develops earlier and the skeletal structures develop temporarily in proximal to distal fashion. This is why the distal structures may, however, develop normally even when more proximal structures develop improperly, providing that the factors causing the proximal anomaly are no longer present during distal limb development. The embryological development of the extremities involves a close association between their ectodermal and mesodermal components. Somatic mesoderm cells migrate to and multiply beneath the surface ectoderm at the limb sites forming the limb buds. After that a discrete specialized zone called the apical ectodermal ridge (AER) forms at the apex of each limb buds. Hemimelia results from a lack of AER-mesodermal interaction during limb outgrowth (Rantanen and Hegreberg, 1982; Towle and Breur, 2004).

Autogenous cancellous bone grafts stimulate osseous union in an arthrodesis (Johnson and Bellenger, 1980; Johnson, 1981). It has been reported that fresh autogenous bone grafts promote new bone formation by introducing osteoblasts and osteogenic precursor cells (osteogenesis), by release of growth factors that induce osteogenic differentiation in mesenchymal tissue at the graft bed (osteoinduction), and by acting as a scaffold on which new bone can be deposited (osteoconduction) (Johnson and Bellenger, 1980; Johnson, 1981). In our case, elbow arthrodesis with autogenous corticocancellous bone grafts resulted in a complete arthrodesis of the joint by 6 weeks of the surgical correction, which is in agreement with the report of Lee at al. (2005).

The failure of the carpal panarthrodesis with metallic plates and screws in this case is thought to be due to screw loosening and instability. The use of acrylic connecting bar in external fixators has become widespread in veterinary orthopedics (Shahar, 2000). It has been reported that a 2 cm diameter acrylic connecting bar is superior to the standard Kirschner stainless-steel external fixator connecting bars in compression and shear loads and performs equally when tested in torsion (Okrasinski et al., 1991). In our case, a 2 cm diameter acrylic connecting bar external fixator was used to perform carpal panarthrodesis which provided good immobilization and stability, and a complete arthrodesis was observed 8 weeks after the surgery. The elbow and carpal arthrodesis resulted in the straightened and corrected the varus deviation of the limb, but still the limb remained shorter than the contralateral limb as we did not perform any decompression surgery to lengthen the limb. However, the surgical corrections resulted in much improvement of the limb function and better quality of life.

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