GUEST EDITORIAL What's New in Limb Lengthening and Deformity Correction

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The Limb Lengthening and Reconstruction Society (LLRS) celebrated its 28th year as an orthopaedic subspecialty, with a combined meeting with the Pediatric Orthopaedic Society of North America (POSNA) at the 2019 American Academy of Orthopaedic Surgeons (AAOS) meeting in Las Vegas. The LLRS 28th Annual Scientific Meeting was held on July 19 and 20, 2019, in Boston, Massachusetts, featuring papers and workshops from around the world. A diverse subspecialty society, we overlap with trauma, pediatric, foot and ankle, sports, arthroplasty, and oncologic orthopaedic surgeons in the diagnosis and treatment of deformities of length, alignment, contracture, nonunion, and bone healing. The purpose of this update was to summarize the important articles with regard to these subjects published since 2018.

Guided Growth

The technique of temporary, asymmetric growth plate tethering continues to be a successful method of deformity correction in growing children. The concept, known as guided growth, was developed by Dr. Peter Stevens, using a small, 2-hole plate to span the growth plate on 1 side, allowing the opposite side to grow unresisted, and thereby correcting angular deformity without osteotomy. The screws within the plate provide an articulation, with the apex of the correction occurring at the perichondrial ring. A multinational retrospective study described results of guided growth in 537 children with 967 physes treated¹. Distal femoral and proximal tibial correction rates were similar, averaging 0.77° per month for the distal part of the femur and 0.79° for the proximal part of the tibia, despite the commonly held opinion that the femur corrects faster than the tibia. Patients with >3 years of growth remaining had a better quality of correction. Infection was uncommon, occurring in 1.78% of patients. Seven of the 8 infections occurred at the upper medial part of the tibia. The study was unable to correlate the effect of body weight on the result, nor did it assess the incidence of rebound deformity.

A modified guided growth technique has been previously advocated for only removing the metaphyseal screw and leaving the plate and epiphyseal screw in place once a correction is complete. Thus, in the event that a deformity recurs, a simpler procedure to reinsert a single screw could be performed. A study by Keshet et al. refuted this practice², as they observed permanent bar formation in 2 of 55 cases using the technique. They also observed that only 12 of their 55 patients required reinsertion of the metaphyseal screw and that 9 of those actually required plate repositioning.

Anterior guided growth of the distal part of the femur to treat fixed knee flexion contracture in cerebral palsy has been reported to have favorable results, provided that implants are placed sufficiently anterior and adequate growth remains³.

Congenital Pseudarthrosis of the Tibia

Congenital pseudarthrosis of the tibia remains a challenge for the orthopaedic surgeon. Richards and Anderson⁴ reported using bone morphogenetic protein (BMP)-2 without deleterious effects in 21 children with congenital pseudarthrosis. They applied a single sponge around an autologous cancellous bone graft with intramedullary pinning that crossed the ankle joint. This technique seemed to shorten the time to union, but did not eliminate nonunion or refracture, the latter occurring in 5 of the 16 initially healed patients.

In a retrospective multicenter study of 119 patients with Crawford type-II congenital pseudarthrosis of the tibia followed to maturity, Shah et al. reported an 86% primary union rate, with 69% still united at maturity⁵. They did not identify specific factors influencing rates of union or refracture and recommended a larger, prospective, multicenter study.

Three-Dimensional Printing

Three-dimensional printing, originally known as additive manufacturing, creates models by printing layers of materials on top of one another. These models can be physically held,

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manipulated, and studied, providing the surgeon preoperative assessment of the specific anatomical geometry.

Haleem and Javaid⁶ described methods to create 3-dimensional models for preparation and practice of orthopaedic procedures. Exact fit models, based on magnetic resonance imaging (MRI) or computed tomography (CT) scanning techniques, can allow for preoperative planning and mock surgical procedures and can create implants specific for complex cases.

Corona et al.⁷ described using 3-dimensional printed titanium truss cages in conjunction with the Masquelet technique to treat massive infected posttraumatic defects.

Bone Defects

Nauth et al.⁸ described a 3-stage protocol of (1) radical debridement and resection of infected skeletal elements; (2) defect space management using antibiotic blocks, spacers, or beads, which provide a local depository of culture-specific antibiotic and develop a vascularized membrane; and (3) 8 weeks later, carefully opening the membrane and exchanging the spacer for an autologous graft.

Napora et al.⁹ described excellent functional outcome of patients at 2 to 8 years after tibial defects were treated with bone transport using a stacked hexapod frame. Corticotomy and transport were delayed for 6 to 8 weeks in patients undergoing concurrent flap coverage. The authors noted improved ease and accuracy of the docking with hexapod frames and described a method to augment frame stability near the end of treatment. The Short Musculoskeletal Function Assessment (SMFA) score was not influenced by age, sex, or diabetic status. Seventeen of 38 patients required an additional fixation surgical procedure (plate, nail, or fixator reapplication) at some point during their treatment.

Barinaga et al.¹⁰ published a case report of successful tibial bone transport combining the use of a motorized magnetic lengthening nail and a limited contact dynamic compression plate, sparing the patient adjunctive external fixation.

Limb Lengthening

Distraction osteogenesis is the spontaneous formation of new bone in a gradually widening osteotomy site using external fixation. The more recent development of motorized internal limb-lengthening nails has advanced the capabilities of treating patients with limb lengthening and reconstruction needs.

Additional authors have reported safe and successful lengthening with motorized intramedullary devices. Iobst et al.¹¹ reported on simultaneously performing distal femoral angular correction and insertion of a retrograde intramedullary lengthening nail, while controlling the fragments with a temporary intraoperative external fixator. Length was subsequently achieved with the motorized intramedullary lengthening nail. Acute corrections of up to 15° were possible with the technique. The authors preoperatively planned their cases using the Baumgart method of reverse planning. Magnetic lengthening nails were used with liberal application of blocking screws preventing postoperative deformity. A cost study suggested that patients undergoing intramedullary lengthening have higher initial implant costs than fixator lengthening over a nail, but the former undergo fewer procedures overall¹².

Reuse of a PRECICE nail (Nuvasive) was reported in a case of a femoral discrepancy after an infection in which a 3-cm lengthening was successfully performed, distal locking screws were removed, the telescopic portion of the nail was retracted, and a second corticotomy and lengthening were performed¹³.

Radiographic evaluation of bone regenerate formation during lengthening is possible by a number of means. A radiographic pilot study by Vulcano et al.¹⁴ found that a pixeldensity ratio of >0.89 (ratio of pixel density in the regenerate relative to the surrounding cortex) may correlate with osseous healing and may be the threshold for allowing full weightbearing. In pediatric patients, Archer et al.¹⁵ also found that the most reliable radiographic assessments of regenerate were by the pixel value ratio and the Ru Li technique.

The mechanical environment of distracted callus affects the quality of new bone formation. An ovine study was performed by Claes et al.¹⁶ in which lateral callus distraction was undertaken in tibiae and various cyclic loads were applied at the end of lengthening. Tibiae that underwent compressive loading had more bone formation, higher spicule height, and higher blood vessel density than those experiencing shear or distractive loading.

Growth Prediction

Makarov et al.¹⁷ evaluated 77 patients at their institution, comparing the predictive accuracy of the White-Menelaus, Anderson-Green, Moseley, and multiplier methods for correctly timing epiphysiodesis. Final leg-length prediction errors varied from 0.7 to 1.1 cm, with the multiplier method proving to be the least accurate. Makarov et al.¹⁸ also reported a 7% complication rate among 863 epiphysiodeses, the most common of which was incomplete arrest in 31 patients resulting in angular deformity and requiring reoperation in half of such cases.

Physeal Violation

A sheep model was used by Knapik et al.¹⁹ to assess the consequences of distal femoral physeal violation by a retrograde intramedullary implant, noting that violations of \leq 7% of the cross-sectional area were associated with continued growth of the physis without bar formation.

Congenital Limb Deficiencies

A review from the Finnish Register of Congenital Malformations and Care Register for Health Care²⁰ identified all children born with lower-limb deficiencies, comparing their hospital admissions and stays with the general pediatric population of Finland. In the 16-year study period, the authors found that children with lower-limb deficiencies (terminal, long bone, The Journal of Bone & Joint Surgery · JBJS.org Volume 101-A · Number 16 · August 21, 2019 What's New in Limb Lengthening and Deformity Correction

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foot, and toe) had 6 times the number of hospital admissions and 10 times the number of days in the hospital per child than all of the children born without limb deficiency. Two-thirds of the patients born with congenital lower-limb deficiencies required an operation, and approximately half of those operations were orthopaedic.

The ratio of the short lower extremity to the contralateral, longer lower extremity in patients with congenital deficiencies has historically been considered constant in the orthopaedic literature. Tsai et al.²¹ attempted to verify this concept of constant inhibition and to further elucidate the pattern of skeletal maturation in patients with fibular hemimelia. They confirmed the concept of constant inhibition, with a mean deviation ranging from 0.1% to 3.3% in the femur and 0.1% to 3% in the tibia. They also noted that the growth patterns of the lower extremities in patients with unilateral fibular hemimelia did not differ when compared with the normal population.

Kowalczyk and Kuźnik-Buziewicz²² reported outcomes of rotation-plasty for unilateral proximal femoral focal deficiency, reviewing postoperative function, time to knee fusion, and complications in 8 adolescents followed for 6 to 12 years postoperatively. Intensive preoperative physical therapy was performed to maximize strength and motion, especially ankle dorsiflexion. Patients were all pain-free and were able to walk without assistive devices. The 2 patients who did not exhibit a Trendelenburg gait had undergone preoperative hip stabilization procedures. Spontaneous derotation of the foot occurred in 5 of 8 patients, necessitating a repeat surgical procedure in 2 patients. All patients reported acceptance of the limb appearance and satisfaction with the result.

Radhakrishna et al.²³ reviewed 6 patients with Jones type-II tibial hemimelia presenting with an absent distal part of the tibia who were treated with limb salvage. All patients were reconstructed with the following elements: gradually moving the fibula distally using an external fixator to normalize the relationship of the proximal parts of the tibia and fibula; centralizing the fibula with creation of a synostosis between the fibula and tibia; and centralizing the foot relative to the fibula distally, creating a fibular-talar fusion, using external fixation and soft-tissue releases. The mean increase in leg length was 4 cm, with a mean time of 43 days in the fixator. Each child required 2 to 4 procedures, with all patients achieving union.

Walker et al.²⁴ analyzed a group of patients with ulnar deficiency and noted that a lower-extremity deficiency was present in 55% of patients. The patients with lower-extremity deficiencies and ulnar deficiencies had less severe presentation of ulnar deficiency. The authors observed no patients with tibial hemimelia (pre-axial deficiency) associated with ulnar deficiencies (post-axial deficiency). They pointed out that the upper-limb bud forms earlier than the lower-limb bud, considering the possibility that a developing limb insult would explain the observation that more lower-extremity involvement was related to less upper-extremity involvement.

Physeal Arrest and Epiphysiodesis

Physeal bar formation was assessed in a rat model by Wattenbarger et al.²⁵. They selectively induced injury by scraping and drilling the metaphyseal and epiphyseal sides of the physis. Basement plate penetration on the epiphyseal side was associated with radiographic and histologic physeal injury, whereas hypertrophic zone violation on the metaphyseal side was associated with continued growth.

Arthroscopic and navigation-assisted physeal bar resection is being increasingly reported. Miyamura et al.²⁶ reported a case with a 2-year follow-up of a distal radial bar excision and concomitant osteotomy, planned using 3-dimensional CT, patientspecific guides, and intraoperative endoscopic visualization.

Foot and Ankle

Ankle arthrodesis for neuropathic foot deformity is historically associated with poor union rates. A fusion technique combining an external fixator and an intramedullary nail was reported to achieve fusion in 22 of 24 patients by El-Mowafi et al.²⁷.

Neuropathic foot deformity with concurrent osteomyelitis is the subject of an article by Kliushin et al.²⁸. Seventy-seven patients were treated for a mean time of 180 days in an external fixator, with infection eradicated in 90.9% of patients at a 1year follow-up. The American Orthopaedic Foot & Ankle Score (AOFAS) increased marginally, which the authors attributed to the pain component of the outcome score. Brandão et al.²⁹ discussed the management of diabetic neuropathy of the foot with both internal and external fixation techniques, with particular attention to the concept of beaming of the midfoot with axial internal fixation. This concept was described as stiff, thick, intramedullary longitudinal support from hindfoot to forefoot.

Staged distraction osteogenesis and ankle arthrodesis were performed by Lou et al.³⁰ in 12 patients with posttraumatic or infectious bone loss of the distal part of the tibia who were treated with simultaneous proximal lengthening and distal compression osteosynthesis using circular fixation to provide ankle fusion. The AOFAS improved from a mean of 37.3 to 75.3 points, and successful regenerate was formed in all, although 2 patients underwent bone-grafting.

Ahmad et al.³¹ reported the results of the treatment of rigid equinus foot deformity with circular external fixation in an adolescent and pediatric population. They reported a mean correction angle of 47.4°. Pin-track infections occurred in 4 of the 30 patients. Equinus deformity secondary to burn contractures treated with circular external fixation was the subject of a report by Zhang et al.³². The authors described a wire technique in the foot, predominantly using hindfoot wires, recognizing the need to avoid injury to the posterior tibial neurovascular bundle.

A cadaver study by Kelly et al.³³ identified safe hindfoot wire insertion using an entry point one-third of the distance along a line from the posterior aspect of the calcaneus to the lateral malleolus to predictably avoid the posteromedial neurovascular bundle. The Journal of Bone & Joint Surgery • JBJS.org Volume 101-A • Number 16 • August 21, 2019 What's New in Limb Lengthening and Deformity Correction

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Nepalese children who were 1 to 5 years of age and had neglected clubfoot deformity were treated by the Ponseti method³⁴. Ninety-five percent of feet remained plantigrade at a minimum of 10 years. Despite some residual deformity, patient-reported outcomes were satisfactory, with a relapse rate of 3%.

Trauma

The role of external fixation in the management of tibial fractures was described in a number of publications. In a retrospective study, Berven et al.³⁵ compared Ilizarov frame fixation with locking plate fixation of proximal tibial fractures between 62 patients treated at 1 center using the Ilizarov technique and 68 patients treated at a different institution using internal fixation. Time to union was considered faster in the internal fixation group. Deep infection, alignment, reoperation, and range of motion were similar in the 2 groups. Lovisetti et al.³⁶ reported the results of circular external fixation in bicondylar tibial plateau fractures, with a mean follow-up period of 37.3 months. All 20 cases were initially managed with knee-spanning fixation. The results were measured by radiographs and Hospital for Special Surgery (HSS) score and were described as favorable when compared with other series presented in the literature.

Napora et al.⁹ examined functional outcomes in 38 patients with infected tibial nonunion managed with stacked hexapod external fixators for the purpose of bone transport. Corticotomy and transport were delayed for 6 to 8 weeks in patients undergoing concurrent flap coverage. The study noted that, with an intact fibula, the proximal and distal portions of the frame programs must be a mirror image of each other and that if angular, translational, rotation, or additional length deformity correction is built into the frame, a fibular osteotomy is required. Patients completed an SMFA survey at the time of final follow-up. The SMFA score was not influenced by age, sex, or diabetic status. Of 38 patients, 17 required an additional fixation surgical procedure (plate, nail, or fixator reapplication) at some point during their treatment.

Arsoy et al.³⁷ reported that patients presumed to have aseptic nonunion but later determined to have positive cultures had an 84% union rate with systemic antibiotic therapy.

Tumor

Elalfy et al.³⁸ determined that chemotherapy had a negative effect on bone healing and diminished bone hypertrophy when using modulated compliant compressive forces to induce osteointegration in a tumor prosthesis. The 49 patients receiving chemotherapy postoperatively had less bone formation than the 44 patients who did not receive chemotherapy. Implant

survival was not affected by chemotherapy, with 10-year survival rates of 85%.

Limb-salvage techniques in the management of pediatric sarcoma continue to evolve. Segmental metadiaphyseal distal femoral defects after resection can be reconstructed with a novel vascularized fibular A-frame technique, augmented with an intramedullary nail. This provides a biological reconstruction with greater cross-sectional area than a single vascularized fibula, with a segment amenable to later limb lengthening³⁹.

For patients with limb growth potential remaining who require endoprosthetic reconstruction, growing prostheses are described. These include both minimally invasive devices (requiring lengthening under fluoroscopic guidance) and non-invasive devices (driven by an external magnet), and these were compared in a survival analysis by Medellin et al.⁴⁰. Prosthesis survival was demonstrated as 79% (minimally invasive) and 70% (noninvasive) at 10 years. Patients receiving noninvasive prostheses had a higher mean Musculoskeletal Tumor Society Score (27 points) compared with patients who received minimally invasive devices (24.7 points), which, although not attaining significance (p = 0.295), was attributed by the authors to higher patient satisfaction, better gait, and greater independence.

A novel approach to limb salvage in sarcoma of the distal part of the tibia was also described by Lou et al.³⁰, with 5 patients undergoing resection of tumor (including the distal part of the tibia and the talar chondral surface) and subsequent tibial bone transport to achieve tibiotalar fusion. The segment was plated 1 month following the transport phase to expedite frame removal, with mean final Musculoskeletal Tumor Society Score of 88 points.

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