

SPECIALTY UPDATE

What's New in Limb Lengthening and Deformity Correction

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Limb reconstruction surgery is an emerging subspecialty. In the past year, 5 major events highlighted the growing interest in this field. The first combined meeting of the 3 international societies devoted to the study and advancement of limb lengthening and reconstruction—the Association for the Study and Application of the Methods of Ilizarov (ASAMI) International, the Limb Lengthening and Reconstruction Society (LLRS)-North America, and the International LLRS (ILLRS)—was held in November 2015, in Miami. Second, a new journal, the *Journal of Limb Lengthening & Reconstruction*, was launched. Third, the *Limb Lengthening and Reconstruction Surgery Case Atlas* was published (Springer International; S.R.R. and R.C.H., editors). This is a major work in association with the LLRS, with 3 volumes and 302 cases covering all aspects of limb reconstruction. Fourth, the LLRS supported and participated in the AAOS/OREF/ORS (American Academy of Orthopaedic Surgeons/Orthopaedic Research and Education Foundation/Orthopaedic Research Society) Clinician Scholar Career Development Program. Lastly, the LLRS Traveling Fellowship program was implemented.

In addition to these events, numerous articles related to limb reconstruction and deformity correction were published in 2015. This Specialty Update provides a summary of this recent research.

Pediatrics

Congenital Limb Deficiencies

Several studies investigated the treatment of congenital limb deficiencies. Shahcheraghi and Javid found that the majority of children with tibial hemimelia had a good functional outcome following reconstructive procedures or knee disarticulation¹. In

a study involving the largest series of tibial hemimelia cases reported from a single institution, other investigators used a modified Jones classification that included a “type V” for cases with global deficiency of the tibia. There were no cases of type III². In a study involving children >4 years of age with type-II fibular hemimelia, Popkov et al. concluded that tibial lengthening should be associated with resection of the fibular anlage in order to prevent recurrence of the valgus deformity of the ankle³. On the basis of radiographic assessment of the feet of patients with fibular hemimelia, Reyes et al. noted that the deficiency “should not be viewed as a global ‘lateral lower-limb deficiency’ nor the foot ray deficiency as ‘lateral.’”⁴ Other investigators found satisfactory results following a specific protocol of lengthening in children with congenital femoral deficiency⁵. Osteonecrosis of the femoral head in the contralateral hip was described for 3 children with congenital femoral deficiency⁶. Prophylactic femoral rodding with Rush rods following fixator removal was reported to be a safe and effective method to decrease the incidence of fracture following lengthening⁷. However, femoral lengthening with circular external fixation had a higher rate of complications than that with use of a motorized nail⁸.

Congenital Pseudarthrosis of the Tibia (CPT)

Three studies provided additional insight into the treatment of CPT in pediatric patients. Comparable results were found between those with and without neurofibromatosis type I who were treated with use of the same Ilizarov technique⁹. Refracture was the main issue following Ilizarov bone transport¹⁰. Proximal tibial lengthening after initial union of CPT was an effective form of management of tibial shortening¹¹.

Blount Disease

For patients with Blount disease who require minimal lengthening, correction using the Taylor Spatial Frame (Smith & Nephew)

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can potentially be performed safely without osteotomy and fixation of the fibula¹². A new classification system for complications in patients with Blount disease treated with a circular external fixator also was presented¹³.

Skeletal Dysplasia

Several reports addressed lengthening in children with achondroplasia. First, tibial lengthening had a significantly lower complication rate than did femoral lengthening¹⁴. Second, although a complication rate of 70% was reported for children with achondroplasia who underwent lengthening procedures, none of these children were left with any long-term sequelae, and all were pleased with the results¹⁵. Third, although double-level tibial lengthening can be difficult, the results are usually gratifying¹⁶. Finally, humeral lengthening was reported to increase the independence of children with achondroplasia, indicating that it is more than a cosmetic procedure¹⁷.

In addition, the successful use of the Fassier-Duval telescopic rod in the correction of deformity in 4 patients with Stüve-Wiedemann syndrome was reported¹⁸. A treatment protocol for the correction of deformity in patients with skeletal dysplasia involving the use of a circular external fixator was also presented¹⁹.

New Techniques

In children with osteogenesis imperfecta, the nonunion of long bones was successfully treated with compressed sandwich allograft cortical struts²⁰, and unicortical locking-plate fixation was successfully applied as an adjunct to intramedullary rodding in cases of fractures and osteotomies²¹. Ippolito et al. reported on a 2-stage technique for the successful treatment of patients with polyostotic fibrous dysplasia and severe coxa vara²². The use of flexible intramedullary nails augmented by external fixation as an alternative treatment of open metaphyseal diaphyseal tibial fractures in adolescents was reported by Atef and Tantawy, with satisfactory results²³.

A new technique for the resection of a central or mixed physeal bar by a complete transverse osteotomy at the metaphysis was also reported²⁴. In another report, satisfactory results of plate-assisted lengthening of the femur and tibia were found²⁵. No improvement in function was found for 11 patients with Legg-Calvé-Perthes disease or developmental dysplasia of the hip who underwent femoral head reduction osteotomy²⁶. Other investigators described a novel technique for the treatment of chronic patellar sleeve fractures using the Taylor Spatial Frame²⁷. Laine et al. reported on the management of 8 patients with open fractures of the tibia with bone loss²⁸. The technique included static stabilization for soft-tissue healing and acute shortening, with a plan for subsequent limb lengthening. Ulnar osteotomy and gradual correction using the Ilizarov external fixator was described for 4 cases of chronic anterior Monteggia lesion²⁹. A successful modified technique for the use of the Taylor Spatial Frame in patients with cubitus

varus after malunion of a pediatric supracondylar fracture was reported³⁰. The risk of proximal rod migration in patients with osteogenesis imperfecta who had deformity correction with use of a Bailey-Dubow rod could be reduced by complete correction of the angular deformity and optimal placement of the rod at the distal physis³¹. Anam et al. reviewed a series of 110 patients with osteogenesis imperfecta who had 261 intramedullary rodding procedures, 139 in the femur and 122 in the tibia, and showed that the introduction of a new medical and surgical protocol for deformity correction (a bisphosphonate infusion-free interval of 4 months after the osteotomy, use of an osteotome instead of a power saw, and the introduction of parenteral zoledronic acid) led to a decrease in the rate of delayed osteotomy healing when compared with the treatment of patients using the old regimen³². Shore et al. compared the results, cost, and complications of 42 children with diaphyseal tibial fracture treated with use of a Taylor Spatial Frame or a uniplanar fixator and reported equivalent cost for care delivery by using either fixator³³. In patients who underwent a transtibial amputation, osteocartilaginous capping with a proximal part of the ipsilateral fibula prevented recurrent overgrowth in 90% of the cases³⁴.

Other Pediatric

The combined use of internal and external osteosynthesis allowed considerable reduction in the duration of the external fixator and decreased the number of complications in children undergoing leg lengthening³⁵ and in children with X-linked hereditary hypophosphatemic rickets who underwent deformity correction³⁶. Preoperative psychological intervention as part of an interdisciplinary team approach was recommended for patients undergoing limb reconstruction³⁷. Another study sought to determine the normal radiographic values of anatomical and mechanical alignment angles of the lower limbs of children of various age groups, the first such chronological evaluation in a relatively large series of patients³⁸. As found in a cadaveric study of femora of donors 8 to 20 years of age by Park et al., nail entry at the pediatric greater trochanter apex would likely result in anterior placement³⁹. Lastly, in cases of poor or delayed bone formation during lengthening procedures, the addition of compressive forces to the distraction protocol (the accordion maneuver) may be beneficial⁴⁰.

Trauma

Hypertrophic Tibial Nonunion

Salih et al., in an attempt to predict which diaphyseal tibial fractures would progress to hypertrophic nonunion, reported on a radiographic sign, termed the *callus fracture sign*⁴¹. This had a sensitivity of 69.6%, specificity of 91.3%, and positive and negative predictive values of 88.9% and 75.0%, respectively. Xu et al. retrospectively reviewed the data of 12 patients with a diagnosis of hypertrophic nonunion treated with an Ilizarov ring fixator, with a union rate of 100%⁴². In no cases did the authors perform compression at the site of nonunion to

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augment healing. In another study, distal tibial hypertrophic nonunions treated with a circular fixator demonstrated union in 100% of the patients⁴³. Patients treated with the Taylor Spatial Frame had improved deformity correction. In a study by Ferreira et al., the authors reported a union rate of 89.1% among 46 patients with aseptic, stiff tibial nonunions treated with hexapod external fixators and distraction⁴⁴.

Tibial Fracture

A meta-analysis compared various modalities for the treatment of open tibial shaft fractures⁴⁵. Method ranking revealed that unreamed nailing was the modality with the highest probability of being the best treatment, followed by reamed nailing, external fixation, and plate fixation. A systematic review of 57 studies evaluating the treatment of open type-III tibial shaft fractures concluded that circular frames have surgical outcomes that are equivalent to those of other techniques⁴⁶. In another systematic review evaluating the outcome of Ilizarov treatment of infected tibial and femoral nonunions, an average 97.26% rate of union was shown for the included studies⁴⁷. In a series of 56 patients treated with use of a Taylor Spatial Frame for tibial shaft fractures, the indications for circular fixation remained inconclusive⁴⁸. Circular fixation remains a powerful tool, especially for cases in which a type-IIIB injury can be converted to a type-IIIA with a technique called *intentional deformation*⁴⁹.

Lengthening and then nailing (LATN) and lengthening and then plating (LAP) are 2 methods of decreasing the time that patients spend in an external fixator. Bernstein et al. reported on 58 patients with posttraumatic bone loss (mean, 5.3 cm) treated with either classic circular fixation and bone transport or “integrated fixation” (LATN or LAP)⁵⁰. They demonstrated a 100% union rate and no difference in the severity or number of complications between the 2 groups. In a study involving an animal model of tibial fracture, there was a subclinical degeneration of myelin fibers in the peroneal and tibial nerves during fracture healing and following fixator removal⁵¹.

Distal Tibial/Pilon Fracture

In a randomized comparative study, 40 patients were treated with open reduction and internal fixation with conventional plating or Ilizarov circular fixation⁵². The rate of healing in the Ilizarov fixation group was higher ($p = 0.003$). The addition of ankle arthroscopy for visualization of the articular surface did not show additional benefit for 23 patients with closed pilon fractures treated with Ilizarov external fixation⁵³. In a retrospective case series of 28 open pilon fractures, Danoff et al. reported on their protocol of the use of staged open reduction of the articular surface and Ilizarov fixation⁵⁴.

Femoral and Tibial Bone Transport

Massive bone loss in the tibia and femur was reported in a study of 110 patients with infected nonunion treated by bone

transport⁵⁵. Ilizarov circular fixation was used in the tibia, and a monolateral rail was used in the femur. The mean bone defect, post-debridement, was 6.15 cm. The authors reported a 100% union rate, with no recurrence of infection. The successful use of a monolateral external fixator was demonstrated in another study of 7 patients with a mean infected femoral bone defect of 8.1 cm⁵⁶.

Other Trauma

The use of circular fixation with classic stainless steel Ilizarov rings has been demonstrated to be safe and cost-effective in austere and low socioeconomic settings⁵⁷⁻⁵⁹. Tibial plateau fractures, Schatzker type-IV to VI fractures, can be effectively treated with Ilizarov circular fixation⁶⁰. In a retrospective study of 177 patients, the rate of venous thromboembolism in patients with tibial fractures treated with circular fixation was 4% (deep-vein thrombosis, 1.7%, and pulmonary embolism, 2.3%)⁶¹. Zhong et al. reported on their single-stage reconstruction for severe foot and ankle deformities that included the use of a sural neurocutaneous flap, noting no flap failures or neurovascular complications⁶².

Adult

Rozbruch et al. retrospectively examined data regarding surgical cases and outpatient visits from the limb-lengthening and complex reconstruction service at one institution⁶³. Referrals were primarily from orthopaedic surgeons (56%) and self/Internet research (25%).

Limb Realignment and Arthritis

There were several studies that supported the use of realignment osteotomy to offload the arthritic compartment of a knee with osteoarthritis⁶⁴⁻⁶⁹. Stupina et al. concluded that restoring the microarchitecture of the subchondral bone and improving its vascularization after tunneling with the introduction of bone marrow suspension into the drill-holes enhances chondrocyte metabolism and recovers their functionality⁷⁰. The success of open-wedge osteotomy is impacted by the completion and speed of bone growth in the triangular space created at surgery. As found in one systematic review, the lowest rates of delayed union/nonunion were in autograft bone-filled osteotomies⁷¹. While high tibial osteotomy (HTO) is the most common knee realignment procedure performed, distal femoral osteotomy was evaluated in the treatment of 21 knees for valgus knee malalignment with arthritis or to protect a knee compartment in which chondral resurfacing surgery had been performed⁷². Harris et al. studied the outcomes of patients undergoing simultaneous meniscal transplantation, osteotomy, and articular cartilage repair, noting improvements in validated patient-reported clinical outcome scores at long-term follow-up⁷³. Konopka et al. studied the cost-effectiveness of HTO and unicompartmental knee replacement as alternatives to total knee replacement for patients 50 to 60 years of age⁷⁴.

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Limb Lengthening

While bone lengthening with external fixation has been demonstrated to be successful, newer methods of integrated fixation are showing promise for some patients by decreasing external fixator-related complications and the duration of time needed in the frame^{75,76}. Dehghan et al. showed that the transplantation of autologous bone marrow mesenchymal stem cells (BM-MSCs) positively affected early osseous consolidation in distraction osteogenesis⁷⁷. Published results of the use of the PRECICE Intramedullary Limb Lengthening System (Ellipse Technologies) in >250 cases were reported to be excellent, with less pain and lower complication rates than with external fixation methods or previous implantable nail systems⁷⁸. Patients who had experienced both internal and external fixation preferred the internal lengthening device with respect to overall satisfaction⁷⁹. The lengthening nail might be superior to external fixation for femoral lengthening, when the anatomical conditions and the complexity of the deformity allow the use of an intramedullary nail^{80,81}. A low-cost custom knee device with polyester synthetic conformable casting material for the treatment of knee flexion contractures was used successfully in 23 patients (27 limbs) who developed knee flexion contracture during femoral lengthening with an intramedullary lengthening femoral nail⁸². Elbatrawy and Ragab studied the outcomes of 50 patients who underwent stature lengthening for psychological reasons and concluded that the Ilizarov device is a safe tool for limb lengthening in individuals of short stature⁸³.

Limb Alignment and Osseous Anatomy

Zampogna et al. compared the measurement of the anatomical femorotibial angle on a knee radiograph with the measurement of the mechanical axis on a full-length standing hip-to-ankle radiograph⁸⁴. The knee radiograph measurement had weak correlation with the long radiograph. In addition, as concluded by Lee et al., alignment after total knee arthroplasty should be assessed by measuring mechanical alignment on a full-length standing radiograph rather than by anatomical alignment⁸⁵. In another study, by Cho et al., the foot progression angle and the mechanical axis of the lower limb were significantly correlated with the first and second peak knee adduction moments⁸⁶. Weinberg et al. studied the association between rotational deformities of the femur and tibia and the development of arthritis using an osteological collection⁸⁷. As assessed by multiple regression analysis, neither tibial torsion nor femoral version proved to be independent predictors of hip or knee arthritis. Weinberg and Liu found that increasing tibial length relative to femoral length was a significant predictor of ipsilateral hip and knee arthritis⁸⁸.

Deformity Correction

A deficiency of the lateral compartment of the knee, often in the setting of skeletal dysplasia, is an intra-articular deformity resulting in genu valgum. Advancement of the lateral femoral condyle can be performed to correct the deformity⁸⁹. Alexis et al. evaluated the utility of the Taylor Spatial Frame among cases treated in Haiti⁹⁰. Good results were observed in 99% of the cases.

Symmetrical distribution of the load of the lower limbs and balance are among the determinants of proper biomechanics of the musculoskeletal system. In a study of 57 patients, load distribution of the lower limbs in the study group after realignment surgery did not significantly differ from that in normal controls⁹¹. The same group examined 56 patients who underwent derotational corticotomies of the distal epiphysis of the femur or the proximal epiphysis of the tibia using the Ilizarov method⁹². The authors found no differences between the study group and controls in physical and sports activity after surgical correction.

Limb Salvage and Amputation

Circular external fixation may be used to stabilize limbs in an intentionally short position and then gradually correct the deformity^{93,94}. The overall incidence, prevalence, fall characteristics, and risk factors were calculated for falls in an amputee population resulting in rehospitalization⁹⁵. Life-threatening infections can occur while using circular external fixation⁹⁶. A satisfactory outcome was obtained with early diagnosis and aggressive medical and surgical treatment.

Other Adult-Related

The Return to Run (RTR) clinical pathway, which was introduced in 2009, has demonstrated favorable results among returning active-duty service members in terms of running, sports participation, active duty, and deployments⁹⁷. Rivera et al. hypothesized that nonsteroidal anti-inflammatory drugs (NSAIDs) may be delivered locally in a wound for the potential prevention of heterotopic ossification⁹⁸.

Foot and Ankle

The use of fluoroscopy was recommended while using an Ilizarov semicircular external fixator for distraction osteogenesis in brachymetatarsia⁹⁹. A study of complex ankle arthrodesis showed a high fusion rate with minimal complications, confirming the utility of ringed fixators¹⁰⁰. A review of the literature on complex ankle arthrodesis yielded an algorithm for surgical treatment¹⁰¹. For a patient with complete fibular agenesis, Cavadas and Thione successfully used the proximal part of the contralateral fibula including the growth plate to craft a lateral malleolus with microvascular anastomosis¹⁰². In a study by Lohia et al. on resistant idiopathic clubfoot in toddlers, both gradual correction with external fixation and open release with acute correction were found to provide significant improvement¹⁰³. A group of plastic surgeons treated foot ulcers associated with Charcot arthropathy by several methods, including free tissue transfer, concluding that aggressive treatment can yield a high healing rate¹⁰⁴.

Additional Studies of Interest

In 4 cases of elbow arthrodesis, circular external fixation was successful at providing a stable and functional upper extremity when amputation was the alternative¹⁰⁵. Three studies showed creative uses for Ilizarov mini-fixators in the correction of hand contractures: a severe intrinsic-plus hand deformity was treated

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with selective releases combined with fixator-assisted stretching¹⁰⁶; carpometacarpal (CMC) arthritis with associated metacarpophalangeal (MCP) joint hyperextension deformity was treated with CMC arthroplasty and metacarpal osteotomy and external fixation with excellent results¹⁰⁷; and, in the final study, postoperative rehabilitation with the use of rubber bands was emphasized¹⁰⁸. A study of 7 patients treated with resection and bone transport using a classic Ilizarov circular fixator yielded excellent or good results in all cases¹⁰⁹. A larger series of 38 transports included classic ring transport with multiple osteotomy sites and Ilizarov crossed olive-wire transport and showed excellent results for the entire group after an average lengthening of 10 cm¹¹⁰. Alzahrani et al. investigated the role of sclerostin antibody infusions in a mouse model of tibial fracture-healing and reported rapid healing when compared with controls¹¹¹. A recent review of distraction osteogenesis and methods for improving bone regeneration focused on the use of adipose stem cells. The study outlined the relative abundance of cells in the adipose tissue and the equivalent performance of these cells when compared with crest-derived cells¹¹². Improved delivery of bone morphogenetic protein (BMP) and other potent bone-healing stimulators may be possible with the use of a chitosan sponge, where the BMP is released more evenly and over a sustained period¹¹³. A case of a 10-year-old patient with anterior cruciate ligament (ACL) reconstruction-related growth arrest and subsequent deformity showed successful use of the lengthening and plating technique¹¹⁴.

In a study involving patients who had tibial surgery and external fixation, Lundblad et al. used PET/CT (positron emission tomography/computed tomography) bone scans to measure bone activity and showed that several static measurements could be used as a proxy for a complete dynamic scan, thus saving examination time¹¹⁵. An investigation of distraction osteogenesis in a sheep model focused on the callus production rate, volume, and mechanical parameters¹¹⁶. In a study of distraction osteogenesis involving dogs, investigators found that the concentrations of growth factors (GFs), including insulin-like GFs 1 and 2, stem cell factor, vascular endothelial GF, and transforming GF- β 1, varied with the rate of distraction¹¹⁷.

A novel tensioning and clamping system that resists loss of wire tension far better than traditional clamping constructs was evaluated by Bairaktari et al.¹¹⁸. Striving for dynamization, Capanni et al. found that dynamic locking plates (utilizing a silicone interface between plate and screws, providing elastically suspended locking holes) behave more like an Ilizarov fixator than a traditional locking plate¹¹⁹. Additional insight into the dynamic properties of circular fixation through sequential testing of various

frame constructs was provided by Henderson et al. Their hypothesis, that half-pins would create a cantilever system that increased the presence of undesirable strain at the fracture site, was disproved by data showing that the all-half-pin construct demonstrated less strain than the all-wire gold standard¹²⁰. Oblique fractures have presented challenges to surgeons using external fixation. Looking at this problem in detail, Jabbar and Khaleel found that placing crossed olive wires across oblique fractures resulted in the most stable constructs¹²¹. Using a transverse fracture and osteotomy model, researchers found that a hexapod-type (Ortho SUV) frame, which allowed for a dynamized mode, was better able to resist deforming forces than was a classic Ilizarov external fixator¹²².

Several other articles were dedicated to answering practical questions regarding relevant human anatomy. The method of vascularized free-tissue transfer was further refined by the use of perforating vessels in place of the larger, named vessels at the anastomosis site¹²³. The authors found no significant increase in complications, despite connecting to a reduced-flow vessel. The use of external fixation around the distal part of the femur creates concern for knee-joint contamination and septic arthritis. A cadaveric study demonstrated that the safe corridor for extra-articular fixation is narrower than perhaps previously thought¹²⁴. Iatrogenic injury of the common peroneal nerve during wire placement in the fibular head is a devastating complication of the application of circular fixation. An anatomical study was conducted among cadavers to precisely outline the safe zone for proximal fibular transfixation¹²⁵. Ktistakis et al. analyzed 369 articles on the topic of pin-site infection after external fixation and concluded that there is no clearly superior method for conducting pin care¹²⁶.

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