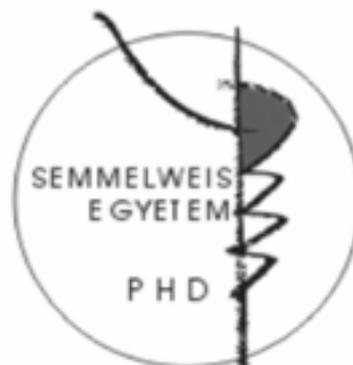


# Special use of fixateur externe in the treatment of developmental musculoskeletal disorders and bone tumours

PhD. thesis

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## **Introduction**

The advance of external fixators in the last decades, and the novel understanding of leg lengthening permits of reasonable correction of congenital and acquired deformities. However in nowadays several difficulties and unanswered questions exist. I present the analyses, results and conclusions of five topics in the dissertation.

With children suffering from fibular hemimelia significant limb length discrepancy can develop, for which the treatment of limb lengthening is carried out most frequently at the affected side, while on the opposite side epiphyseodesis is made. For the adequate treatment the most accurate assessment of the final length of the lower limbs is required. With children suffering from fibula hemimelia on the basis of our earlier experiences we have observed the change of the bone maturing, which made the assessment of the maturing and the final limb length inaccurate.

Our knowledge on the cause and prevention of the joint contracture, one of the most frequent complication of limb lengthening, is insufficient even today. During femoral lengthening most often knee joint flexion contracture develops, while during tibial lengthening the emerging of equinus contracture is characteristic.

Humeral lengthening in comparison to femoral or tibial lengthening is very rare. One of the factors possibly explaining the rarity of humeral lengthening may be that a minor shortening of the humerus rarely constitutes either a functional or cosmetic handicap to the patient. Only a few reports on lengthening of the humerus can be found in the body of international orthopaedic literature, while reports on humeral shortening are more prevalent. The development of the callotasis method and improved lengthening apparatus has provided us with the technology to successfully correct length discrepancies of the upper extremity.

The features of exostosis cartilaginea multiplex are osteochondromas, which are at present simultaneously on more bones, starting mainly from the metaphysis of the long bone of the limbs. The discrepancy appears with a frequency of 80 % on the distal part of the ulna, and causes the shortening of the ulna, the bending of the radius and the ulnar direction slip of the proximal carpal bones in the radiocarpal joint. The classification of the forearm deformities caused by exostosis on the ulna and the radius has been made by Masada on the basis of the morphology features.

A significant part of the malignant tumours of the lower limb start from the proximal metaphysis of the tibia. In adolescent and mature age the most frequent procedure is the resection of the tumour and the supplement of the missing segment by tumour prosthesis. During the childhood, however, the open physal plate functions as border of compartment from the point of view of tumour spread, thus the epiphysis can remain intact. This characteristics enables radical tumour resection in the height of the growing plate, as well as the retention of the tibial condyles. For the supplement of the removed bone segment fibula transplantation is preferred above all, as most often a section of 10-15 cm must be bridged.

## **Purposes**

- 1.** The purpose of the study in the patients' group with fibula hemimelia was to establish a nomogram by sequential determining of the skeletal age in order to predict the expectable limb length more correctly.
- 2.** The purpose of the study with animal model was to get to know the effect of leg lengthening of different rates and sizes on the change of the range of motion of the surrounding joints. We had also wondered as to whether the age influences the development of the contracture.
- 3.** The aim of this study was to examine the effectiveness of lengthening the humerus in children and young adults.
- 4.** The purpose of the study was to assess the treatment results of the forearm curve caused by the multiplex osteochondromas by lengthening the shortened ulna.

5. Our purpose was to assess the new method for treating the malignant tumours starting from proximal metaphysis of the tibia, which was made by retaining the epiphysis and the autologous fibula transposition, and an Ilizarov circular fixator for fixation was used.

## **Methods**

### **1. Errors of prediction of limb length based on the Moseley table in fibular hemimelia**

In thirty-one children with unilateral fibular hemimelia the skeletal maturity was monitored by analyzing data collected on skeletal age and limb length. At least four (on the average seven) X-ray pictures were made on each child, and minimum one year passed between the X-ray pictures in each case. On making the first X-ray picture the average age of the patients was 5 years and 4 months (between 3 years 4 months and 6 years 8 months). On making the last X-ray picture the average age of the patients was 19 years and 5 months (between 9 years 4 months and 27 years 2 months). Out of the 31 patients 26 were monitored until the skeletal maturity. Five patients still did not reach the skeletal maturity until the end of the study, their average age is 11 years 8 months (between 9 years 4 months and 13 years 2 months). The length of the limb and the skeletal age was determined 6.8 times on the average during maturity with each patient. We made the X-ray pictures on the bones by standard technology. We determined the skeletal age by comparing the antero-posterior X-ray picture made on the left wrist and the skeletal maturity atlas of Greulich and Pyle. Each data were put in the Moseley nomogram in order to create a new nomogram. We applied linear regression for the sake of a function best adjusted on the points.

### **2. Change of range of motion of the malleolar joint during tibial distraction osteogenesis in animal model**

We chose 16 young and 12 matured rabbits to the various experiment groups. We lengthened the tibia of the rabbits by external fixator, depending on the group, with different distraction rates. On the entities of the group serving as the elongation control (n=3) after mounting the osteotomy and the fixator no lengthening was made. We lengthened the left back limb of the animals grouped in all experiment group, whereas the right back limb served as control. The animals were killed directly after the end of the lengthening procedure. The rate of lengthening was, depending on the group in which the animal was drawn, 0,8 mm/day, 1,6 mm/day and 3,2 mm/day, respectively. The size of lengthening was 20%, 30%, and 40% of the original length of the tibia in the groups, respectively. For measuring the motion of the ankle joint we prepared a joint ankle meter, the flat sheet of which was fitted to the sole of the rabbit, whereas the thin metal bar on the other side of the equipment was placed on the tibia. We read the angle values from the angle meter fastened to the sole sheet. The measuring could be repeated by 5 degrees accuracy. 4-6 measuring were made on each joint, the average of which was regarded as the result. Each measuring was made directly after killing. As control we regarded the whole extension+flexion range of motion of the ankle joint not lengthened, the value of which was 170° on the average (80°+90°, from the full extension to full flexion).

### **3. Principles and conclusions of humeral lengthening**

Fourteen humeri (twelve patients; six males and six females) were elongated using the Wagner fixator. The average age of the patients at the time of surgery was 17.2 years (range: 12–32). The average amount of shortening was 8.4 cm (range: 4.5–13.5 cm). The most common reason for humeral shortening was congenital in origin in this group of patients. The lengthening was performed with Wagner midi or normal equipment. Following insertion of the fixator, we sought and carefully isolated the radial nerve, followed by osteotomy in the middle of the humeral diaphysis. The lengthening protocol was a 7-day latency period, followed by 0.5-mm twice daily distraction. The patients learned how to handle the fixator, to take care of the area surrounding the pins and the exercises. The patients were discharged on postoperative day 12–14, and the lengthening procedure was continued in their homes.

Range-of-motion tests at the elbow and shoulder were carried out every 2 to 4 weeks, and the axis of the humerus and bony remodelling were checked every 2 to 6 weeks. We left the fixator in the humerus until total bony reconstruction was complete, and there was no need for plate fixation or bone transplantation.

#### **4. Forearm deformity correction by ulnar lengthening in exostosis cartilaginea multiplex**

We made ulna lengthening in seven cases because of lower arm shortening and radius axle discrepancy which developed on exostosis cartilaginea multiplex. With six patients the exostosis of the ulna had been chiselled off earlier, with one patient this was made together with the placement of the fixator. Out of seven patients in 3 cases the ulna was lengthened with circular Ilizarov equipment, whereas with 4 patients by linear Wagner equipment. During the operation first we placed – depending on the type of the external fixator – a készüléket a csonthoz rögzítő Kirschner drótokat vagy menetes szegeket into the proximalis and distalis part of the ulna. After the placement of the lengthening equipment we made osteotomia on the ulna meta/diaphysis, then we applied compression between the osteotomised bones for 7 days. Following these we started the lengthening of the ulna by a rate of 1 mm/day. The lengthening lasted until reaching the length of the ulna in ratio with the radius length. We left the equipment on the limb until sufficient bone healing demonstrable by radiology, then it was removed. During the lengthening and also afterwards we paid special attention to the retention of the motion of the elbow and the wrist. The age of the patients changed between 5.9 and 13.4 years at the time of starting the elongation, it was 11.5 years on the average. All operations were made before the end of the skeletal maturity. The follow up time was 1.7-5.9 years, 3.9 years on the average. During this period the maturity was completed with three patients. We evaluated on X-ray pictures the shortening of the ulna, the ulnaris tilt of the distalis radialis joint surface, as well as the slipping of the carpal bones.

#### **5. Epiphysis saving procedure in proximal tibial tumour**

Between the years 2002 and 2007 143 patients were treated with malignus lower limb tumour, out of which in 29 cases hip, in 63 knee-, in 13 diaphysealis tumortprothesis implantation and in 38 cases bone graft supplement was carried out during the limb retention operations. With five patients the tumour started from the proximal metaphysis of the tibia, but did not affect the proximal epiphysis. With these patients the tumour resection was in the growing cartilage, thus we preserved the tibia epiphysis. The youngest patient was 5 years old, the eldest 14, their average age was 11.2 years. The histology examination proved osteosarcoma with two patients and Ewing-sarcoma with three. All of the patients received neoadjuvant chemotherapy. We determined the size of the tumours and the tumour free state of the epiphysis by MR pictures. This operation was made if the proximal tumour spread did not reach within 2 cm of the growing cartilage. With four patients we excised the tumour proximalis in the growing cartilage, with one patient 2 cm under the physis. We had to remove the tuberositas tibiae together with the metaphysis with one patient. The distalis resection level changed according to the size of the tumour, this was mainly in the middle of the diaphysis. For the supplement of the bone insufficiency we used both the opposite side and the identical side fibula. The fibula from the other side was transplanted free together with the fibula head in a way that the head was shaped cubicle by sawing off the cortical parts. We mobilised the fibula head of the identical side, then by creating spongiosus surfaces they were also transplanted into the epiphysis. We cut the fibula under the resection level of the tibia and fixed it with wire cerclage to the tibia. We fixed the limb with an Ilizarov equipment, in which we placed a ring in the tibial epiphysis, one to the distalis femur metaphysis and two to the distal tibia. We permitted full loading, which was reached by the patients gradually in 2-20 weeks. The proximal ring stabilizing the knee joint was removed after 18-20 months. The whole equipment was removed after 16-25 (average 21.2) months. At

this time first we applied plaster cast fixation for 6-8 weeks, then provided the patients with a free knee joint orthosis. The postoperative chemotherapy was conducted according to the usual procedure. We have been following up our cases for 3.9 years on the average (2.5-7 years).

## **Results**

### **1. Errors of prediction of limb length based on the Moseley table in fibular hemimelia**

We illustrated the average data of the patients suffering from unilateral fibula hemimelia on the Moseley-type normal skeletal maturity nomogram in order to create a new nomogram particularly for this patient group, also displaying the standard deviation. The skeletal age data of this patient group differ from the scheme of the skeletal maturation process of the healthy population. Between the ages of four and six the average skeletal age of a sick child falls behind that of the healthy children. From the age of six until the skeletal maturity the skeletal maturation process of the sick children accelerates compared to that of the healthy children. As a consequence of the above two statements the curves illustrating the skeletal age of the sick children and that of the healthy children must cross each other. The two axes cross each other around the age of 10.5 with girls, whereas around the age of 12 with boys. After the crossing, the nomogram prepared by us shows further decrease, consequently the sick children reach the skeletal maturation earlier than their healthy fellows. The angle closed by the horizontal line in the Moseley-type nomogram and the monotonously decreasing lines in the new nomogram is 31 degrees with girls and 33 with boys. The skeletal maturation scheme of the patients suffering from unilateral fibula hemimelia, which differs from the normal, will result in an inaccurate prediction of the future limb length difference on the basis of the Moseley-type nomogram without the correction proposed here. This will appear in an over- or under-correction of the length difference.

### **2. Change of range of motion of the malleolar joint during tibial distraction osteogenesis in animal model**

Evaluating the experiment with animal model we did not find any change in the motion in the control group with osteotomy and fixator implantation but not lengthened. With the matured animals 0.8 mm/day, with 20% lengthening the average range of motion decreased to 129° (to 76%) (25-180°, SD:22,9). In the matured animal group 1.6 mm/day, with 20% lengthening the extension and flexion significantly decreased further on compared to the former group: the total motion to 53° (to 31%) (60-145°, SD:23,3). Young animals 0,8 mm/day, lengthened by a rate of 20% did not show any discrepancies against the control size on measuring. However, the daily elongation of 1.6 mm caused the restriction of movement of moderate degree at the same age, which affected the extension range: range of motion changed to 35° (to 79%) (35-180°, SD:8,2). Similarly, the lengthening of 0,8 mm/day by 30 % of the young animals resulted in a slight decrease of the range of motion: 150° (to 88%). The lengthening of 3.2 mm/day of the young animals caused the development of complete size (90°) equinus contracture, shortly after starting the procedure.

### **3. Principles and conclusions of humeral lengthening**

The average amount of lengthening was 6.7 cm (range: 4.5–10.5 cm), and the proportion of achieved lengthening was 30.4% (range: 16–44%). The average healing index was 32 day/cm (range: 25–40 day/cm). The lengthening procedure was well tolerated by all the patients. The patients' co-operation was favourable, and all could maintain their normal daily activities – with slight restrictions. Pin tract infection was found in every second patient, but after local treatment and oral antibiotics, suppuration stopped. Two temporary elbow flexion contractures developed (20° and 45°, respectively); however, after the contracture had been recognised, physiotherapy was initiated, with a subsequent complete return of the range of motion. One patient had neuropraxis that resolved within 3 months of commencing

physiotherapy. The planned lengthening was achieved in every case. Nonunion or other major complications were not observed.

#### **4. Forearm deformity correction by ulnar lengthening in exostosis cartilaginea multiplex**

Out of 7 patients suffering from osteochondroma, on the basis of the Masada ranking, six had a lower arm deformity of type I., and one patient type II/B. In three cases we made ulna lengthening on the right side, and in four cases on the left side. On the basis of the preoperative X-ray pictures the shortening of the affected ulna changed between 31-60 mm against the opposite side, it was 42.8 mm on the average. This length difference of 15.8-26% equals with a shortening of 20.5% on the average. The duration of the ulna lengthening amounted to 81-209 days (146 days on the average), during which period a lengthening of 18-50 mm, 31.3 mm on the average, could be achieved. This means the lengthening of 10.4-30.3% of the original ulna length, which is 18.6% on the average. The lengthening was carried out in each case until the ulna reached the line of the articular line of the radius. Consequently the lengthened ulna had been shorter also postoperative than at the opposite side. The postoperative shortening changed between 7-20 mm, it was 11.5 mm on the average, this equals relative ulna shortening of 2.8-9.5% (5.8% on the average). The preoperative radius joint angle of 32-50 degrees, 40.7 degrees on the average, decreased to 39.2 degrees postoperative. The sliding of the carpal bones from the range of 75-100% before the start of the lengthening (88.5% on the average) improved to 20-80% (47.1% on the average) by the end of the lengthening.

#### **5. Epiphysis saving procedure in proximal tibial tumour**

Among the patients with tibia tumour resection we did not notice any local recidiva during the observation time, no metastasis developed. We experienced proper fixation of the transplanted fibula with all of the patients both proximal to the tibia epiphysis and distal to diaphysis. The range of motion of the knee was in one case 10-30 degrees, with the others these changes between 0-120 and 140 degrees, 130 degrees on the average. Two patients can walk without support and one in orthosis with full loading of the limb. Two can walk in orthosis with partial loading. The limb shortening of the affected side can be measured 1.5-4.5 cm, 2.6 cm on the average. With three patients the transplanted opposite side fibula has broken. Wearing the Ilizarov-equipment we noticed excretion periodically with all patients along the Kirschner-wires. In one case the Kirschner-wires of the ring placed in the tibia epiphysis broke. With one patient chronic osteomyelitis developed at the distal part of the transplanted fibula.

### **Consequences**

- In unilateral fibula hemimelia we stated different skeletal maturation from those of healthy people, therefore if we use the Moseley nomogram or the Green-Anderson graphs for the prediction of the expectable limb length then we will get an inaccurate result. By sequential determination of the skeletal age we have created a modified nomogram, by which the limb length development can be predicted more correctly.

We examined the effect of limb lengthening of different rates and sizes on the change of the range of motion of the malleolar joint at animal model, and we have demonstrated that

- by increasing the rate of lengthening the danger of the joint movement restriction will grow exponentially.
- A normal rate of distraction does not result in joint movement distraction with young animals, whereas with matured animals contraction will appear.
- A doubled lengthening rate causes with young animals moderate, with mature animals serious joint movement restriction.
- Increasing the size of lengthening, even by normal daily rate, results in a slight decrease of the range of motion with young animals.

We have proved by upper arm lengthening carried out in human practice that

- the unilateral equipment can be well used for lengthening the humerus,

- spongiosaplasty and plate fixation is not necessary,
- lengthening much over 15%, even of 50% can be carried out.

Exostosis cartilaginea multiplex:

- The lower arm deformity caused by ulna shortening of type Masada I and II/b, and the ulnar instability of the radiocarpal joint is not ceased by chiselling off the ulna exostosis.
- The slipping of the carpus can substantially be improved by lengthening the ulna.
- The unilateral equipment of linear type is the proper choice for lengthening.

We have developed a new method for the treatment of malignant tumours starting from the tibial proximal metaphysis,

- which were carried out by retaining the epiphysis and the transposition of the autologous fibula, we applied Ilizarov-equipment as fixator.
- By the treatment we could achieve final limb retention.