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LOWER LIMB LENGTHENING: EXPERIMENTAL STUDIES OF BONE REGENERATION AND LONG-TERM CLINICAL RESULTS

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to my family

to my friends

to my teachers

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ABBREVIATIONS AND TERMINOLOGY

ad libitum – latin...as much as...
AO/ASIF – arbeitgemeinshaft für osteosünthesefragen/association of the study of internal fixation
b.w. – body weight
DE – distraction epiphysiolysis
DXA – dual energy x-ray absorbtiometry, used for measurement of bone density
e.g. latin *exempli gratia* – for example, for instance
EDTA – ethylene diamine tetraacetic acid
EM – electron microscope
L% – lengthening percentage
LI – lengthening index
LL – limb lengthening
ROM – range of movement
MC – metaphyseal corticotomy
MO – metaphyseal osteotomy
OI – *osteogenesis imperfecta*
SFCE – slipped femoral capital epiphysis

Callotasis – Callo from the latin noun *callum* – scar tissue between the fragments destined to transform itself into bone tissue, tasis from the ancient Greek noun *Táois*, meaning tension or extension
– Callotasis is a surgical method that lengthens bone by means of distraction of the callus forming around the osteotomy site after a waiting period (Aldegheri 1993)

Corticotomy (compactotomy) – means a low-energy cortical osteotomy with transection of only the bone cortex. The periosteum, the endosteum, the bone marrow with its blood supply, as well as the muscles and soft tissues surrounding the bone are maximally preserved
Corticotomy metaphyseal vs compactotomy diaphyseal

Healing/lengthening index – used for description of bone lengthening, is counted by dividing the duration of lengthening (days, weeks, months) by the amount of length gained (cm)

Questionnaire – a set of questions submitted to persons receiving health care in order to share their view on the treatment received and perform an outcome evaluation, usually patient-administered. Tool, instrument survey or tool are often used as synonyms

Outcome – the end result of a clinical intervention

1. INTRODUCTION

The process of growth, being most vulnerable to the effects of chronic diseases, nutritional and psychosocial deprivation and adverse environmental conditions, constitutes one of the most sensitive parameters of health (Desai 2003). Limb lengthening procedures are performed to correct significant length discrepancies in the upper or lower extremity that have resulted from various congenital or acquired abnormalities (Walker *et al* 1999).

The issue of limb lengthening has been actual in orthopaedic surgery up to the present time. Shortening of one lower limb affects the vertebral column and causes scoliosis, stiffness of the ankle joint in the shortened leg and stiffness of the knee joint in the normal leg; an altered centre edge angle of the hip joint overloads the femoral head. Thus shortening of the limb by one centimetre evokes a 3.5 degree deviation of the vertebral column (Roesler and Rompe 1972). In the case of 3 cm shortening of one lower leg scoliosis will be 10.5 degrees.

Although experimental studies have proved the effectiveness of different methods of limb lengthening, there is still room for investigation of the mode of regeneration of the bone tissue. The goal is to clear up the role and response of the cortical bone, periost and endost in distraction limb lengthening and in filling of bone defect and their response to these procedures? Are there any important vascular or bone-intrinsic impacts, or even some endocrine factors? Therefore carrying out of experiments on different laboratory animals is still actual.

It seems to be a reasonable approach that manipulations are performed on a deformed shorter limb. Patients strongly desire limb lengthening, despite the higher risk of this operation, and are disinclined for a shortening operation on the contralateral limb. The lengthening operation restores the patient's height, whereas the alternative procedure leaves the patient deformed through disproportionate shortening.

However, there have been several “fashions” in limb lengthening, especially in terms of gaining the total length of the man. There is no need to make a short stature patient a bigger one. It is important to improve the patient’s functional ability and coping. Many investigations have demonstrated the need for teamwork, and the importance of psychological approach. This yields better outcome and satisfaction of patients. On the other hand, there have emerged facilities for cosmetic limb lengthening (Catagni *et al* 2005) and the problem of iatrogenic crippling is arising as described in Lancet (Watts 2004).

Modern tecniques developed in the 1950s and 1960s in Russia and in the 1980s in the Western world resulted in the elaboration of various distraction devices and methods of limb lengthening.

The choice of the method of limb lengthening is still controversial and needs further investigations. If a simultaneous deformity correction requires joint-neighbouring procedures like distraction epiphysiolysis and metaphyseal lengthening are appropriate. It is common that during limb lengthening the angular deformity correction is performed (Birch and Samchukov 2004). Diaphyseal lengthening, based today mainly on callotasis on the intramedullary nail, has some developments for automatic lengthening systems (Hankemeier *et al* 2005). Several traumatic limb lengthening methods (immediate, Wagner), resulting in quite a large number of complications, have been abandoned, but in the case of certain indications they may have some advantages.

A number of studies have shown that one-stage limb lengthening over 20–25% is not good for functional means or in regard to complications. Other studies do not confirm this viewpoint. Yet extensive limb lengthening has yielded good results. Investigations of the reasonable amount of limb lengthening are still actual.

Although the complication rate of limb lengthening is high, not many of them affect the final result. As long-term satisfaction of limb lengthening is not well studied and only some studies have been published (Ghoneem *et al* 1996, Ramaker *et al* 2000, Karlen *et al* 2004), relevant investigations should be continued.

Different methods of limb lengthening do not differ in long-term results, more important factors are age and the type and aetiology of deformity. Patients are quite satisfied despite the fact that some psychological problems arise during the procedure. Patients who underwent limb lengthening because of congenital deformities or for sequelae of osteomyelitis were less satisfied compared with patients with acquired deformities or with posttraumatic patients. Yet most the patients were ready to pass limb lengthening once more if it would be indicated.

The first limb lengthening in Tartu University Hospital was performed in 1976. At the same time, external fixators in the completion of orthopaedic surgery were widely used in Estonia, as well as in Tartu. This experience resulted in academic dissertations on limb lengthening (Haviko 1977, 1989) and fracture treatment (Lenzner 1986, Poljanski 1986).

The present study comprises experimental investigations of bone regeneration, especially the response of bone, periost and endost to distraction limb lengthening. Clinical studies include functional outcome, long-term results and patient satisfaction after lower limb lengthening.

2. REVIEW OF THE LITERATURE

2.1. History of limb lengthening

The history of limb lengthening and external devices has been largely the history of forgotten and newly discovered ideas, as is often the case with other fields of life. Successful lengthening of the extremities was first reported at the beginning of the twentieth century. The reports describing methods such as the distraction of fragments, callus or epiphysis for the first time were reviewed. The publications of Codivilla (1905) introduced lengthening by fragment distraction. Despite the fact that Lambotte developed an external fixator in 1902 (Taillard 1984), Ombredanne was the first to use an external fixator for limb lengthening. In 1913, he reported lengthening of bone at a rate of five millimetres per day after the production of an oblique osteotomy of the femur; however, skin necrosis and infection occurred (Aronson 1997). In 1921, Putti (1921) slowed the rate of distraction to two to three millimetres per day with a monolateral fixator and half-pins (Aronson 1997).

In 1923 Bier reported bilateral femoral lengthening, but he failed to recognize the bone forming potential of the periosteum and the endosteum and hence the need to protect their vascular supply (Wiedemann, 1996). On the other hand, Bier is called the pioneer of callus distraction (Bertram *et al* 1999). In 1944, Wittmoser (Brug *et al* 1991) devised a ring fixator for lengthening the tibia and fibula. This device could have solved all earlier mechanical problems and it resembled the apparatus in current use. His teacher, Lorenz Böhler, however, failed to recognize the brilliance of the underlying idea and could see no link with any earlier studies, least of all those of August Bier. Böhler commented, "Your device looks very pretty, but do not publish it. Otherwise someone might use it and that would be a major disaster." (Brug et al., 1991; Wiedemann, 1996). Pais, in 1946, was probably the first surgeon to carry out distraction along an intramedullary nail followed by Küntscher in the 1950s (Wiedemann, 1996). The principle of direct desmoid ossification had been described in 1937 by Stefan Krompecher (1956). Ilizarov followed these ideas and elaborated the principles of his method which are the following:

- superior biologic quality of the regenerated bone due to the performance of a percutaneous corticotomy that causes minimal trauma to the periosteum and bone marrow
- a postoperative waiting period
- multistep, incremental distraction totalling 1 mm/day
- use of a compression and distraction procedure involving full weightbearing
- use of a ring fixator in which the fragments are held by Kirschner wires under tension, which enables the surgeon to exert planned axial control in all planes and even to correct multidirectional deformities
- development of segment transport for defects of the bone shaft

– promotion of good tissue nutrition and joint mobility by means of a mobile device that allows full weightbearing and physiotherapy (Paterson 1990; Cech and Trc 1993; Wiedemann, 1996, Aronson 1997).

In 1963 Wasserstein (1971) carried out gradual two-stage lengthening along an intramedullary metallic lamellar rod where a cylindrical cortical allograft was used in the second stage to fill the distraction gap created by lengthening (Wasserstein 1971, 1990; Paterson 1990; Wiedemann, 1996). Zavyalov and Plaksin (1968, 1969) were the first to use the technique of distraction epiphysiolysis in patients. DeBastiani and the coauthors (1986a; 1986b) described distraction of the growth plate named chondrodiatasis. In 1983, Monticelli and Spinelli presented a method of metaphyseal limb lengthening. Many patients have been operated using methods of diaphyseal lengthening (Wagner 1971), resulting nowadays in limb lengthening, on an intramedullary telescopic or automatic nail (Bliskunov 1983; Garcia-Cimbrelo *et al* 2002, Hankemeier *et al* 2005).

2.2. Distraction bone healing in experimental investigations

Bone repair and distraction gap healing are specific forms of wound healing of the mesenchymal tissues. They are complicated processes owing to the formation of the provisional metaplastic repair organ the callus. Contemporary limb lengthening surgery is mainly based on the experimental histomorphological study of the callus. Three different modes of ossification during bone lengthening by distraction osteogenesis were found on a rat model. They are endochondral ossification, intramembranous ossification, and transchondroid bone formation (Yasui *et al* 1997; Li *et al* 1999; Choi *et al* 2002).

In comparative studies of different species of laboratory animals, dogs have proved to be the most suitable (Eitel *et al* 1981).The bone ages of humans and rabbits were compared and it was found that one rabbit’s day is equal for fourty human days (Jani 1975). New bone formation during distraction osteogenesis is well organized and spatially isolated from the process of bone resorption. Several studies have demonstrated that the histological pattern of bone formation by distraction osteogenesis in dogs, rabbits, rats, and mice is analogous to that in humans (Aronson 1994; Tay *et al* 1998; Aronson *et al* 2001; Song *et al* 2002).

2.2.1. Experimental bone healing in the case of different operation techniques

Different locations of the bone distraction gap like the diaphysis (Ilizarov and Berko 1980; Ilizarov *et al* 1969; Ledyaev 1975; Shtin *et al* 1979), growth cartilage (Jani 1975; Haviko 1977; Startseva and Gorbunova 1982; Peltonen *et al* 1984ab; DePablos *et al* 1986) and metadiaphyseal region (Ilizarov *et al* 1975; Ilizarov *et al* 1977; Gylnazarova and Shtin 1983; Ilizarov *et al* 1984) have been experimentally investigated. In growth cartilage investigations, distraction epihysiolysis has been used, while in metaphysis investigations osteotomy or corticotomy have been used. Ilizarov and Shreiner (1979) described for the first time corticotomy and closed flexion osteoclasia with transsection of only the bone cortex. The time of the appearance of osteoblasts in the distraction gap was determined after diaphyseal osteotomy of the dog femur and endosteal regeneration was observed on the fifth day at the place of the bone cut. On the 14th day of distraction, activated osteoclasts as well as an increased number of small blood vessels were detected in the distraction gap (Ilizarov and Berko 1980). However, in another investigation, lengthening of the tibia, an increase in the amount of osteoclasts was only seen on the 35th day of distraction (Ilizarov *et al* 1975). The role of osteoclasts was investigated by Chambers (1980). He showed that in the case of activated osteogenesis, the number of osteoclasts increased on the bone trabeculas.

2.2.2. Experimental metaphyseal limb lengthening

Ilizarov and the coworkers (1969; 1977) in their experimental and clinical studies demonstrated the need for a waiting period of 10–14 days after osteotomy or corticotomy. This period is important for development of the primary regenerate between the bone fragments. Optimal waiting time in experimental diaphyseal lengthening was 10 days (Shtin *et al* 1979). In a morphologic study Shtin and Nikitenko (1974) demonstrated that the optimal latency period after corticotomy was 7–10 days. More recent experimental research suggested that the metaphyseal site was the optimal location for distraction osteogenesis. The biological difference in the osteogenic potential between the metaphyseal bone and the diaphyseal bone seems to be significant in bone regeneration and mineralization (Welch *et al* 1998)

Shugarov and Arapov (1975) proved that insertion of the Kirchner wire due to the vibration of the pin tip interrupted circulation inside the bone, which restored only after 30–60 days. Even though the intramedullar vessels were interrupted by osteotomy at surgery, blood circulation recovered during the waiting period before distraction. Based on the results of these experiments and on the clinical experience of 180 bone lengthenings, the authors believed that a waiting period after osteotomy was more reasonable than achieving immediate distraction after uncertain corticotomy (Yasui *et al* 1993).

Steen and Fjeld (1989) proved in their experimental study on sheep that the inferior mechanical stability of the bone segments in metaphyseal compared with diaphyseal lengthening (due to differences in frame rigidity and distribution of muscular moments) influenced healing to such an extent that any superior biological osteogenic potential in the metaphyseal bone region was nullified. Therefore, the empirically accepted idea that metaphyseal healing is superior may not be correct (Steen and Fjeld 1989; Steen *et al* 1990).

2.2.3. Factors influencing bone regeneration

Many investigations have demonstrated good circulation in the forming bone inside the distracion gap (Ilizarov *et al* 1969; Lavrishtsheva and Dubrov 1968; Ledyaev 1975; Pflüger *et al* 1976; Pouliquen *et al* 1980; Dusmuratov and Dusmuratov 1984; Sveshnikov *et al* 1985). Blood supply to normal bone and bone healing are the main regulators of cell proliferation and differentiation between chondroblasts and osteoblasts (Wilson 1991; Remedios 1999). An important factor of bone repair is also the intact periosteum (Landry *et al* 1996; Gordh *et al* 1997). Kojimoto and the coauthors (1988) showed that the periosteum was the most important structure in bone healing followed by intramembraneous ossification whether or not the medullary canal has been disrupted. Earlier Ilizarov and coauthors (1969) demonstrated that bone formation would occur more rapidly and more securely if the endosteal structures were left intact. Experiments on dogs showed that new bone was formed from both the medullary cavity and the periosteum (Hamdy *et al* 1997). Coleman and Scott (1991) pointed out the importance of all structures.

Different influences, e.g. bone resection, perforation, and periostectomy yielded different results (Goransson *et al* 1992; Einhorn 1995). Formation of bone with a large mineral content did not require intactness of bone marrow, however there was an effect of interaction between bone marrow and periosteum on healing (Guichet *et al* 1998). Song and the coauthors (2002), comparing the callus at the bone defect with the callus at the distraction site of simple lengthening, did not find a significant difference in bone quality. Numerous general and local factors of the environment (blood supply, state of osteogenic and –inductive zones) influenced bone healing (Heikkinen *et al* 1974; Grundnes and Reikeras 1991a; Grundnes and Reikeras 1991b; Tuukanen *et al* 1992; Guerino *et al* 1999).

2.3. Lower limb lengthening

2.3.1. Current clinical techniques of limb lengthening

Many investigations have been devoted to different methods and techniques of limb lengthening, while very controversial data have been published.

Corticotomy and callotasis have been the methods of choice in diaphyseal and metaphyseal lengthening. Corticotomy (compactotomy) represents low-energy cortical osteotomy with transsection of only the bone cortex. The periosteum, the endosteum, the bone marrow with its blood supply, as well as the muscles and soft tissues surrounding the bone are maximally preserved (Schwartsman and Schwartsman 1992). The same authors have described the failures of corticotomy: corticotomy is performed like osteotomy but without proper regard for the vascularity of the bone and the surrounding soft tissues, displacement after corticotomy arising from incorrect application of the frame, and incomplete corticotomy with premature closure of the corticotomy site. The way of performance of corticotomy was important (Baumgart *et al* 1997; Yasui *et al* 2000).

Callotasis is a surgical method that lengthens bone by means of distraction of the callus forming around the osteotomy site after a waiting period (Aldegheri 1993).

Different methods of bone cut and the time for starting distraction have been studied. The term callotasis and the theory of callotasis were devised and developed between 1975 and 1985 by Aldegheri (1988, 1989, 1993, 1997), DeBastiani and the coauthors (1987). Alho and the coauthors (1982) used the term osteotaxis distraction before the term callotasis was introduced. However, Ilizarov and the coworkers (1971;1973) in their clinical studies demonstrated the need for a waiting period of 7–14 days after osteotomy or corticotomy.

Even Abbott (1927) and other earlier researchers used delay before distraction, but usually for purposes other (pain, swelling) than waiting for bone regeneration (White and Kenwright 1991). Bier was named the pioneer of callus distraction: he introduced the idea of an about 3–5 – day waiting period after osteotomy (Bertram *et al* 1999). The relationship between the latency periods and corticotomy sites suggested that the latency period for either metaphyseal or diaphyseal lengthening sites may not be necessary (Aronson and Shen 1994).

2.3.2. Methods of operation

Limb lengthening in children by distraction of the growth plate was first described by Zavyalov and Plaksin (1968, 1969) and Ilizarov and Soibelman (1969) who used a technique that they called distraction epiphysiolysis, which invariably resulted in separation of the epiphysis from the metaphysis.

DeBastiani and the coauthors (1986) described a technique for slow, progressive, symmetrical distraction of the growth plate, named chondrodiatasis. Using this technique in 58 children, including 25 achondroplastic patients, lower limbs were lengthened with good results.

More recent clinical and experimental investigations of distraction epiphysiolysis revealed a danger of growth retardation independent of age, but individual variations were considerable and the extent of retardation was not predictable. So the procedure was advocated namely in the age before skeletal maturity (Startseva and Gorbunova 1982, Bjerkreim 1989, Fjeld and Steen 1990).

Franke and the coauthors (1990) compared distraction epiphysiolysis and partial metaphyseal corticotomy, and analysed their advantages and disadvantages. They concluded that the main goal of metaphyseal lengthening was to avoid premature closure of the growth cartilage and the procedure can be done throughout childhood. After lengthening, the epiphyseal cartilage started to function in all patients, and premature growth cartilage fusion was not observed during follow-up. The authors suggested that physeal distraction is a good method for moderate and simple limb lengthening. Particular care should be applied to knee joint function, especially during femoral lengthening (Haviko 1977; Zarzycki *et al* 2002). In tibial lengthening the function of the ankle should be observed; in the case of equinus deformity Achilles lengthening should be performed (Aldegheri 1999). The placement of pins of the external fixator was important to the function of knee joint (Simpson and Barker 2002). Still good results have been obtained using physeal distraction (Langlois and Laville 2005). Some authors report growth disorders even after the use of the callotasis method (Sabharwal *et al* 2000).

Growth rate was drastically reduced after 30% or more of lengthening using the callotasis technique and in 50% of cases there was evidence of a premature closure of the distal femoral growth plate. Caution should be exercised in carrying out high percentages of lengthening of children's bones (Lee *et al* 2001).

Metaphyseal lengthening was preferred to diaphyseal lengthening since metaphyseal bone is expected to heal faster than diaphyseal bone (Fischgrund *et al* 1994). Monticelli and Spinelli (1983) presented a method of metaphyseal distraction lengthening which can be used after closure of the epiphyses. They showed that the method was more reliable, quicker and had fewer complications due to the greater osteogenesis potential of the metaphysis compared to the diaphysis. A significant difference (especially in lengthenings less than 7 cm) was found between metaphyseal and diaphyseal lengthenings. More intensive osteogenesis and greater mechanical strength have been described as the two advantages of metaphyseal lengthening (Moseley 1989). White and Kenwright (1991) concluded that the level of osteotomy is located at the junction between the metaphysis and the diaphysis, an area with a greater potential for bone healing than the mid-diaphysis. In cases of short stature double site, so-called bifocal limb lengthening have been used (Saleh and Hamer 1993; Kristiansen and Steen 2002). Special attention should be paid to soft tissue because it did not lengthen independently in bifocal procedure (Aarnes *et al* 2002). Stiffness of the knee joint in femoral lengthening was lesser when corticotomy was performed in the subtrochanteric region (Hankemeier *et al* 2004). Another study showed that metaphyseal osteotomy had superior bone healing compared with diaphyseal osteotomy (Bowen *et al* 1993).

Korzinek and the coauthors (1990) stated that corticotomy at the metaphyseal level was generally successful, whereas in the diaphysis the same procedure was done without a major disruption of the intramedullary circulation and did not affect bone regeneration in only 30% of cases.

2.4. Clinical outcome of lower limb lengthening

2.4.1. Objective indicators of clinical outcome

2.4.1.1. Lengthening index

The healing/lengthening index, used for the description of bone lengthening (Monticelli and Spinelli 1981; DeBastiani *et al* 1987), is calculated by dividing the duration of lengthening (days, weeks, months) by the amount of the length gained (cm), but Sakurakichi and the coauthors (2002) advocated to use distraction, maturation, and external fixator indices separately. They demonstrated that athough the procedure involved almost the same rate of distraction, the waiting period and the maturation period were different, which influenced the total period of lengthening. The healing/lengthening index was influenced by many factors: aetiology of limb shortening; the techniques used like distraction epiphysiolysis, chondrodiatasis, corticotomy/osteotomy and callotasis; the site of the procedure like metaphysis, diaphysis or growth cartilage, the upper or the lower part of the distracted segment; the amount of lengthening; single or double level procedure. Very different values of healing/lengthening indices have been obtained which vary from one month to two months, while usually the value of the index was within one month in double level limb lengthening (Maffulli *et al* 1996; Stanitski *et al* 1996; Aldegheri 1997; Aronson 1997; Noonan *et al* 1998; Yun *et al* 2000; Sakurakichi *et al* 2002; *et al* Zarzycki *et al* 2002; Eralp *et al* 2004; Karlen *et al* 2004). On the other hand, osteogenesis started already in the distraction period and in more extensive lengthenings the healing/lengthening index was usually lesser (Kristiansen and Steen 2002; Sakurakichi *et al* 2002). Besides the above described findings, a negative hyperbolic relationship has been found between the healing/lengthening index and the amount of lengthening in centimetres and percentages. The opinion that healing/lengthening indices may be useful only as a rough estimate of the duration of treatment required for each procedure has been expressed. It seemed more reliable to calculate the time required to lengthen a lower limb segment by one percent (Noonan *et al* 1998).

2.4.1.2. Complications in limb lengthening

The rate of complication in limb lengthening depends on different factors: their grading, the type of deformity, and the amount of limb lengthening. In a study by Dahl and the coauthors (1994), all unwanted events during and after treatment were considered complications and were simply graded as minor and major. The rate of major complications was 72%. Complication rate appears to relate to severity of the deformity rather than to the device used. Complications have been graded as minor, serious and severe, and their appearance has been related to severety of the deformity (Roesler and Rompe 1972). Paley (1990) classified unwanted events into problems, obstacles and complications and believed that subdivision of the difficulties and complications of lengthening describes them more reliably and truly. Dividing complications into major and minor was believed to describe them sufficiently. Major complications included appearance of pseudoarthrosis, delayed regeneration, fracture or deformity of the regenerate, ankylosis or subluxation of the joint and osteomyelitis. Minor complications were: minimal loss of lengthening, nonfixed equinus, superficial infection around pins, transient signs of nerve stretching and swelling. The rate of major complications was 20.6% and that of minor complications was 10.6%, altogether 30.2% (Haviko 1989).

Complication rates have been different when the amounts and methods of limb lengthening were compared, ranging from 30% (Garcia-Cimbrelo *et al* 1992) even to 1.2 (120%) major complications per patient. (Guidera *et al* 1991). Limb lengthening requires careful planning and meticulous outpatient care and is still burdened by minor complications. Experience may lessen major complications, but pin tract problems, swelling, and pain will plague these patients (Eldridge and Bell 1991). Major complication rates during limb lengthening were plotted in a consecutive series to produce a learning curve. There was a significant decrease in complications as experience was gained. Directed formal study and surgical instruction diminished these complications (Garcia-Cimbrelo *et al* 1992; Dahl *et al* 1994). Usually the final result is influenced by a minor part of complications only (Vizkelety and Marschalko 1993) and patients are very happy with the result (O'Beirne *et al* 1993).

2.4.1.3. Complications in large-amount limb lengthenings

Caution should be exercised in carrying out high percentages of lengthening of children's bones (Lee *et al* 2001). Other authors agree that the amount of lengthening with an acceptable complication rate should not exceed 25% of initial bone length. The prevalence of major complications seems to be correlated with the complexity and duration of treatment (Maffuli and Fixsen 1996; Hantes *et al* 2001). In a retrospective clinical study of 111 limb lengthenings by the use of three different operation methods, accounting for 88%, 97% and 100% of the initial length of the segment, the rate of complications was 40% (Korzinek *et al* 1990). In response to historical guidelines, suggesting limits to the amount of limb lengthening, the results and complications of these patients were reported in whom the initial goal of lengthening exceeded 20% of initial segment length. All of these patients had problems and 0.9 (90%) complications per segment lengthened were observed, but good to excellent results were achieved in 78% of the cases (Yun *et al* 2000).

2.4.2. Follow-up in limb lengthening

The most common method used to monitor limb lengthening is plain radiography, but the time of frame removal is still left to the judgement of the surgeon (Hughes *et al* 1994). Interpretation is, however, based upon subjective parameters that have never been clearly defined (Donnan *et al* 2002). Many studies deal with radiographic evaluation of regenerative processes during limb lengthening, but there is no commonly accepted pattern (Fischenko *et al* 1976; Young *et al* 1990; Kolbeck *et al* 1999). Also there are possibilities of ultrasonography, computed tomography and bone densitometry (Hughes *et al* 1994). DXA scan seems to give valuable information about the time of removal of the distraction frame (Reichel *et al* 1998). Thirty-five calluses formed during limb lengthening were classified radiographically into 6 types: external, straight, attenuated, opposite, pillar, and agenetic. Healing indices correlated well with the intrinsic periosteal and endosteal conditions of each type. This classification enabled the authors to estimate intrinsic conditions, predict the healing index, control daily lengthening speed, and decide about application of early augmentation of the callus (Hamanishi *et al* 1992). In the radiographic imaging of lengthened limbs after Ilizarov, it was important to centre plain x-ray films accurately at the distraction site and align the connecting rods outside the distraction gap. A combination of plain radiographs and ultrasound could be utilized to follow the progress of the distraction gap, especially in children (Blane *et al* 1991). Distraction monitoring via osteocalcin together with radiological assessment was advocated, especially for patients with unpredictable bone formation, or if prolonged osteogenesis was already obvious at the beginning of distraction (Fink *et al* 2002).

2.4.3. Long-term results in limb lengthening

2.4.3.1. Electromyographic and function studies in limb lengthening

Surgical lengthening of limbs often results in loss of the range of joint movement. In most cases, the bone lengthening operation did not improve walking, especially because it was associated with serious loss of muscle strength and the author stated, "Function must be regarded as the essence of the matter" (Moore, 1941). Experimental investigation showed that changes in the connective tissue component are important factors in loss of joint movement. In the case of the muscle distracted at a high rate, failure of the muscle fibres to add on sufficient sarcomeres, combined with changes in the connective tissue, resulted in almost total loss of joint movement (Williams *et al* 1999). In 25 patients who underwent isolated Ilizarov femoral lengthenings (mean lengthening, 6 cm), no correlation was noted between the worst ROM (during lengthening) and final ROM at the last follow-up examination (Herzenberg *et al* 1994). The unexpected loss of movement observed in the prelengthening period indicates that efforts of physical therapy must be directed to this phase in order to accelerate the recovery of the joint range and to reduce the muscle-related complications that can occur during limb lengthening (Barker *et al* 2001). Distraction during a limb-lengthening procedure caused substantial morphologic changes in muscles and nerves (Battiston *et al* 1992; Chandler *et al* 1988; Stephens 1983; Haviko 1989). Symptoms from such an injury to peripheral nerves and muscles, although rather common, usually disappeared when the velocity of the distraction was reduced, or after the procedure (Galardi *et al* 1990; Faber et al 1991; Karger *et al* 1993; Rajacich *et al* 1992). However, Sofield et al (1958), Kawamura et al (1968), and Macnicol and Catto (1982) reported gait disturbances and a loss of strength as many as twenty years after a limb lengthening procedure. In an experimental study in rabbits the incidence of neuromuscular injury was common. These findings suggested that early detection of neuromuscular involvement by electrophysiological test is a practical way to assess neuromuscular function during limb lengthening (Chuang *et al* 1995). On the other hand, in the electromyographic testing of 9 patients after bilateral limb lengthening in achondroplasia, only small changes in muscle strength were found postoperatively.

2.4.3.2. Outcome and satisfaction studies in limb lengthening

Treatment of limb length inequality is a complicated and long-term procedure and we must carefully equalize limb length in a manner that is neither physically nor emotionally scarring (Guidera *et al* 1995). Brockway and Fowler already stated (1942), "Leg lengthening will always entail certain hazards and there will probably be some poor results, but with the experience gained we feel that the hazards and poor results can be reduced to a creditable minimum." They gained good or satisfactory results in 87% of limb lengthening patients.

In earlier studies Merle d'Aubigne and Dubousset (1971) gave a description of the grades of satisfaction. It was good when the desired length of the limb was achieved, satisfactory when ¾ of limb length with good function was achieved, and poor when limb lengthening failed or severe restrictions of function appeared.

Psychological and social impact studies of the Ilizarov leg lengthening procedure concluded that nearly all patients and parents were positive about the procedure and the support that they received. Most patients, 88%, stated that they would undergo the treatment again, if it were indicated (Ramaker *et al* 2000). According to another study, 82% of patients were satisfied with the overall result, but only 67% of children thought that the duration of the treatment was reasonable and said that they would be willing to have the procedure a second time if it were indicated (Ghoneem *et al* 1996). Sixteen of 22 patients completed a questionnaire after leg lengthening according to Wagner. Most noted both improved function and cosmesis. The answer to the question "Would you do it again?" was "yes" in 11 and "no" in 5 cases. Eight of the 22 patients did no experience any psychological problem (Hrutkay and Eilert 1990). In comparison with the Ortofix and Ilizarov devices, the scars were shorter and cosmetically better in the Ilizarov group, but the patients in the Orthofix group were more satisfied because they had fewer scars. Patients were also more satisfied with the thigh scars than with the leg scars because clothing can easily cover the thigh scars (Karlen *et al* 2004).

In a study of Tjerneström and Rehnberg (1994) 60 patients were satisfied with the results of lengthening. Only five patients, all with traumatic shortenings, were dissatisfied. Complete patient satisfaction was achieved in 45 of 48 patients (94%) and partial satisfaction was achieved in the remaining three cases (Paley 1990).

2.5. Ollier's disease and limb lengthening

In 1900 L.Ollier, a French professor of orthopaedic surgery and one of founders of French orthopaedics first described the condition known today as Ollier's disease (Little 1994). Ollier's disease (dyschondroplasia, multiple enchondromatosis) is a rare non-hereditary disorder of unknown pathogenesis. Dyschondroplasia is a developmental affection rather than a neoplastic disease (Tachdjian 1990). It is characterized by multiple, asymmetrically distributed, intraosseous cartilaginous foci and subperiosteal deposition of the cartilage with a common pattern of unilateral involvement of the lower extremities. The femur, tibia, and ilium are the bones most often involved. Also, the phalanges and the metatarsals are frequently involved (Bessler *et al* 1992). The tumours are located in the epiphysis and in the adjacent parts of the metaphysis and the shaft. Deformities resulting from tumours include shortening caused by lack of epiphyseal growth, broadening of the metaphyses, and bowing of the long bones. An occasional lesion has characteristics of both fibrous dysplasia and enchondromatosis (Carnesale 2003).

A significant shortening of the limb always causes gait disturbances due to massive and inconvenient orthosis, and leads to a decline in working capacity even up to development of disability. Therefore, correction of extensive limb shortening remains a topical problem in trentment of this pathology (Bonnard *et al* 1993; Glorion *et al* 1995; Kristiansen and Steen 2002; Paley 1990; Saleh and Goonatillake 1995). Despite the long treatment time and usually a complicated course, patients are satisfied with the results even if the cosmetic appearance is not the best. They do well socio-professionally (Dutoit *et al* 1990; Stanitski *et al* 1995).

Different options in treating Ollier's disease have been used starting with amputation (Mitchell and Ackerman 1987) in suspected malignancy with a severe deformity of the limb due to intraosseous cartilaginous foci. Limb lengthening (Tachdjian 1990), either gradual bone grafting (Urist 1989) or callus distraction (Pandey *et al* 1995), were the most common methods of correcting deformities in the lower limb.

Successful treatment depends on efficient interdisciplinary efforts of health care professionals from medicine, nursing, physical and occupational therapy, recreational therapy, and social services. Long-term consideration must address the potential for malignant changes (Little 1994; Tachdjian 1990).

The reviewed literature presented controversial experimental data. The method of osteotomy and the cellular pattern of bone regeneration are still under discussion. A question arose about the role of the periost and endost, about the mode of their co-existence and about their mutual influence. The data of electromyographic studies are controversial in terms of the extent and duration of muscle damage (temporary or permanent). Posttraumatic limb lengthening is believed to heal faster because of nonaffected bone metabolism, however specific analysis of this group is not available. Limb lengthening in Ollier's disease is well studied and replacement of cartilage foci is a known procedure as well, but extensive distraction limb lengthening with continuous follow-up has not been described. While only a few studies are devoted to the problems of satisfation after lower limb lengthening, long-term analysis has not been undertaken.

3. AIMS OF THE STUDY

The aims of the sudy were:

1. To investigate bone regeneration and appearance of cellular composition in rabbit tibia after corticotomy and distraction lengthening (publ I)
2. To investigate the response of the bone, periost and endost in rat tibia after resection osteotomy or perforation and in the case of isolation of the periost (publ II)
3. To study the effect of lengthening of the femur on the extensors of the knee (publ III)
4. To assess treatment of posttraumatic shortening and correction of deformities of the lower limb using the Ilizarov distraction device (publ IV)
5. To evaluate long-term result of extensive limb lengthening in Ollier's disease (publ V)
6. To evaluate the results of lower limb lengthening and patient satisfaction in long-term follow-up (publ IV, V, VI)

4. MATERIALS AND METHODS

Table 1. Experimental material and patients; TU – Tartu University

Study type	Study site	No of patients/ expermental animals	Goals of the study	Publication
Experimental, prospective	TU	20 rabbits	Distraction bone healing after corticotomy of tibia	Acta et Commentationes Universitatis Tartuensis, 1989; 835: 79–88
Experimental, prospective	TU	142 rats	Role of periosteal and endosteal ossification in filling a tibial defect or a perforation hole	Annales Chirurgiae et Gynaecologiae 2001; 90: 271–9
Clinical, prospective, electromyo-graphy	TU	7 patients	Assessment of the knee extensor muscles after extensive femur lengthening	J Bone Joint Surg 1995; 77A(2): 247–50
Clinical, retrospective	TU	14 patients	Treatment of posttraumatic limb shortening with the Ilizarov distraction device	Annales Chirurgiae et Gynaecologiae 2000; 89: 303–7
Clinical, case report	TU	1 patient	Evaluation of extensive limb lengthening in Ollier's disease	Medicina 2005; 41(10): 861–6
Clinical retrospective, questionnaire	TU	53 patients	Long-term results and patient satisfaction after lower limb lengthening	"Outcome and satisfaction studies in limb lengthening" International Orthopaedics, 2006 (submitted)

4.1. Experimental investigations

4.1.1. Corticotomy of rabbit tibia

Experiments were carried out in 20 male Shinshilla rabbits aged 1.5–2.0 months. The ethical guidelines for the care and use of the animals were followed. Lengthening of the right tibia on the level of the lower metaphysis was performed. The left leg was used as control. Histological changes in the distraction gap in different stages of lengthening were investigated. The idea of choosing rabbits was to obtain larger amounts of regenerative tissue, while the rabbit is the smallest laboratory animal available for performing corticotomy.

For anaesthesia, ketamine and diazepam were used. Intramuscularly, 2–3 ml (100–150mg) ketamine and 1 ml (5mg) diazepam were injected, after which sufficient anaesthesia was achieved for surgery.

The tibia was discovered in its lower third on the level of the metaphysis. At first *retinaculum extensorum* was cut and at the same level corticotomy of the metaphysis was performed, special attention was paid not to damage the endost and intraosseal circulaton. A modified mini ring fixator with a ring diametre of 30 mm and with two pairs of crossing pins, one pair through the upper and the other through the lower metaphysis, was set onto the leg. This type of fixator provides sufficient stability as described in the studies of Ilizarov and the coauthors (Ilizarov *et al.*, 1969). Damage to the nutritional artery of the diaphysis was avoided.

The animals were divided into six groups according to the length of the experiment (Table 2): 5 days – 4 rabbits, 7 days – 3, 10 days – 4, 14 days – 3, 21 days – 4, 30–35 days – 2. Distraction was started 2 days after surgery at a rate of 0.35 mm per day.

Table 2. Experimental animals (rabbits)

No experimental animal	4	3	4	3	4	2
Distraction days	5	7	10	14	21	30–35
Distraction mm	1.5	2.3	3.0	3.5	7.2	10.0

4.1.2. Resection and perforation of tibia in rats

In the experiment 142 male growing Wistar rats (200–220 g) were used. The guidelines for the care and use of the animals were approved by the Ethical Committee of the University of Tartu. Rats were used to get more reliable data for comparision of different influences on bone regeneration.

The animals were divided into 3 groups: 1) internal artificial fracture of the tibia (34 rats); 2) osteotomy (40 rats) and 3) perforation of the cortex tibiae

(68 rats). The first group was not divided further. The second group was subdivided into: 1) control only with resection of tibia (18); 2) animals trained by the swimming (12) and 3) immobilized animals (10). The third group was subdivided into: 1) the subgroup with the intact periost (53) and 2) the subgroup with the isolated periost of the tibia (15) (Fig 1). The animals were removed from the study between 1 and 42 days after the operation.

In the present study the first group (34) animals were not analysed. In the second group analysis of 18 control animal with resection of tibia was performed. In the third group filling of the perforation hole with the bone tissue in the isolated periost (15) and in the intact periost (25) of the rats tibia was investigated (Fig. 1, bold boxes)

Anaesthesia was introduced with an intramuscular injection of ketamine 50mg/kg b.w. and diazepam 5mg/kg. Prophylaxis of infection was carried out with ampicillin 7.5mg/kg i.m. an it was started 2 hours before the operation and lasted 3 days.

Operative technique

Resection osteotomy with a length of 4 mm, was performed by sawing below the epipyseal line, between the diaphysis and the proximal epiphysis. The bony fragment was transsected and extirpated. Fibre fixation was performed by a fibula.

A perforation hole with a diameter of 1.3–1.5 mm on the anterior surface of the tibia was drilled through the bone cortex between the diaphysis and the proximal epiphysis. The injury was exactly located and fixed *in situ*, and additional fixation was unnecessary. Isolation of the periost from the endosteal tissues (15 animals) was achieved by covering the perforation opening with medical wax at 37–40ºC. These experiments permitted to ensure: 1) correct and exact dosage of injury and 2) separate assessment of the periosteal and endosteal callus zones, especially in the area of angiogenesis.

The rats were kept in a special box during the postoperative period, 3 animals in each. Special rat food and water were given *ad libitum*.

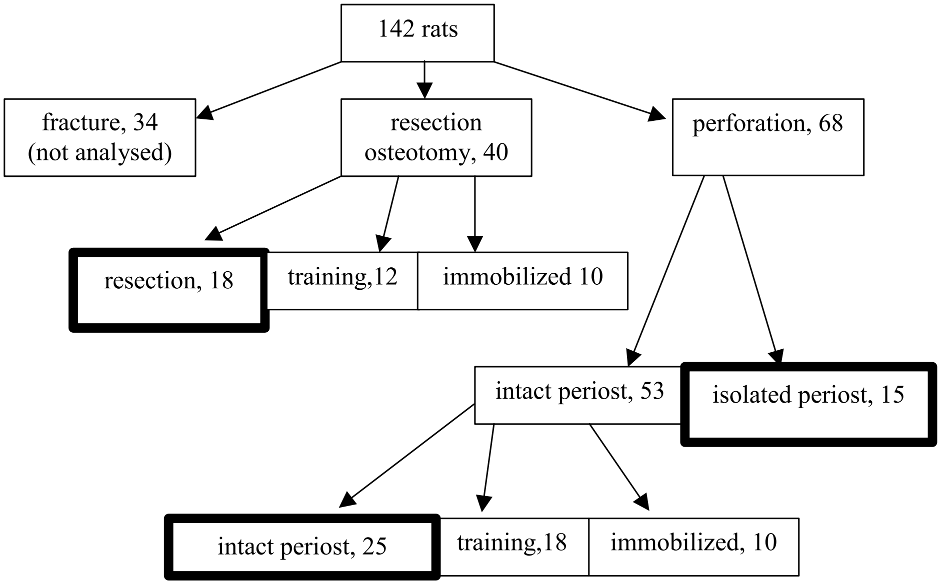


Figure 1. Distribution of experimental animals between the study groups. The groups marked in bold are the study subjects of this thesis.

4.1.3. Histological investigations

The removal of an animal from the experiment was carried out under anaesthesia. For histological investigation, 3 cm of the distal parts of the rabbits tibia were taken; in rats the average size of the portions was 0.5–1.0 cm. The material was fixed with formalin and Zenker formol according to Maximov and demineralized with the solution of Shampy or with EDTA. Paraffin embedded slices with a thickness of 7 µm were stained with haematoxylin and eosin, azure 2 – eosin, Heidenhain iron hematoxylin, and with alcian blue as well as after van Gieson. Microscopically, the main charateristics of the epiphyseal cartilage and the distraction gap were given. After that, hyperaemia was estimated in the regeneration area (the percentage of erythrocyte-filled small vessels from a total of 50 vessels), in the region of newly formed bone, osteoclasts were counted (25 fields ob.40 oc 7) and the bone to connective tissue ratio was calculated. The mitotic activity of the cells was calculated by immersion magnification on the preparations stained according to Feulgen.

Standard electron microscopy was used. A 3% glutaraldehyde solution was employed for fixation and a 1% osmium tetroxide solution was used for contrast enhancing (“staining”). The pH of fixation was around 7.2. Embedding in epon or araldite was carried out. Ultrathin (thickness about 50–100 nm) and semithin sections (0.5–1 μm) were used. Semithin sections were stained with toluidine blue.

The microanatomical images of the callus were copied and saved electronically. Further the process was performed according to the computer program Adobe Photoshop simultaneously with control by light microscopy.

The above pictures were analysed observing the areas of the osseous, chondrous, connective and degenerative as well as inflammatory tissues. Different tissues were painted with Adobe Photoshop in different colours. The painted areas of different colour were summarized in pixels and the proportions of different tissues in the callus area were calculated in percentage.

Thus posttraumatic bone repair was studied histologically (histological slides, EM) and histomorphometrically (histology + computer analysis).

4.2. Patients (clinical analysis)

4.2.1. Distraction limb lengthening

Limb length discrepancy was measured clinically and by plain x-ray technique taking into consideration magnification. The indications for limb lengthening were lower limb shortening more than 3 cm or a severe deformity at the joint level caused by different aetiological factors.

The Ilizarov type of external devices were used. The patient was in a lying or slightly semisupine position depending on surgery in the tibial or femoral segment of the limb. All operations were performed in general anaesthesia. In some cases of tibial lengthening, to avoid equinus contracture in the ankle joint, the Achilles tendon was lengthened in the amount of proposed tibial lengthening using Z-plasty. Then the two rings were connected with threaded rods and the apparatus was set over the leg. The wires used, 1.5–2.0 mm in diameter, were introduced by a power tool at slow speed, while the soft tissue was penetrated without drilling. The entry and exit points of the wires were managed on the extension side of the limb at flexion and in the flexion side at extension. Finally, three or four rings with tightened cross K-wires were set on.

Four types of limb lengthenings were used.

1. Diaphyseal osteotomy was performed by means of the Gigli saw and using the original Wasserstein (1971) method: gradual two-stage lengthening along an intramedullary metallic lamellar rod was carried out where a cylindrical cortical allograft was used in the second stage to fill the distraction gap. The outcome in patients operated by the use of this method, was analysed in 4 cases in an electromyographic study, in 4 cases in a posttraumatic limb shortening study and in one case of tibial lengthening in Ollier's disease.

2. In distraction epiphysiolysis care was taken in introduction of the wires through the epiphysis. Usually an image intensifier was used.
3. Metaphyseal osteotomy or corticotomy was done by using a chisel and a curved osteotome. An effort was made to perform corticotomy at the metaphyseal-diaphyseal junction.
4. Double site tibia lengthening in achondroplasty patients: lower epiphysiolysis and upper corticotomy or corticotomy at both ends of the bone were performed. In four cases double level and single level osteotomies in two patients were performed.

The data of the patients of limb lengthening from groups 2, 3, 4 are presented in Table 3.

Table 3. Data of the 53 patients with epimetaphyseal lengthening

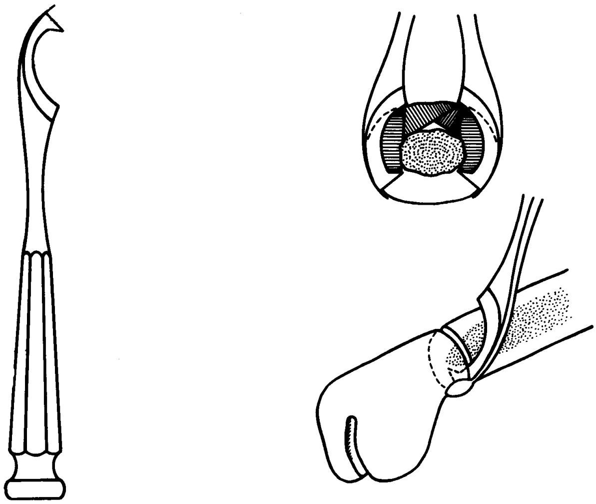
Case	Year	Age	Sex	Dgn	Segment	Method	Ach	Shortening (cm)	Lenghtening (cm)	+/–	Lenghtening (%)	Index	Response
1.	1976	7	m	2	1	1		5	5	0	16.1	23.4	+
2.	1977	14	m	2	2	1		5	5	0	11.9	20.2	
3.	1978	13	f	2	2	1		6	6	0	20.7	36.2	
4.	1978	15	m	3	1	2		22	22	0	75.9	22.5	+
5.	1979	14	m	2	2	1		3	3	0	8.5	20.0	+
6.	1979	10	f	5	1	2		6	6	0	18.8	27.5	+
7.	1979	18	m	5	1	2		4	4	0	8.7	23.5	
8.*	1980	8	f	2	1	3		15	5	–10	20.0	20.2	+
9.	1980	11	f	4	1	3		2	3	1	12.5	10.0	
10.	1981	9	m	2	2	1	+	4	5	1	14.3	35.2	+
11.	1981	8	m	2	2	1		1	3	2	3****	14.0	+
12.	1981	19	f	4	1	2		5	5.5	0.5	16.7	26.5	+
13.	1981	6	f	4	1	2		5	6.5	1.5	23.0	26.5	+
14.	1982	20	m	5	2	3	+	3	4	1	10.5	35.8	+
15.*	1982	18	m	1	2	2	+		7.5	7.5	40.0	22.5	+
16.	1982	8	m	4	1	2		8	9	1	29.0	12.9	
17.	1983	4	f	2	2	2	+	4.5	5	0.5	25.0	26.6	+
18.	1983	12	f	2	2	2	+	4	5	1	16.1	43.8	+
19.*	1983	6	m	1	2	1			13	13	81.0	33.8	
20.	1983	9	f	3	2	1	+	4	5	1	17.5	21.2	+
21.	1983	31	f	2	1	3		7	6	–1	18.2	37.3	+
22.	1983	13	m	4	1	2		5	6	1	14.3	22.8	+
23.	1984	17	f	5	2	2		1	1	0	4****	22.0	+
24.*	1984	8	f	1	2	1	+		11	11	73.3	22.8	+
25.	1984	22	m	2	1	3		5	5	0	11.4	48.8	
26.	1984	8	f	2	1	2		5	6	1	20.0	27.0	+
27.	1984	9	f	4	1	2		5	9	4*****	26.5	25.1	+
28.	1984	13	f	4	1	2		5	5	0	11.9	26.2	
29.**	1984	5	f	5	1	2		5	5	0	17.2	26.4	+

Table 3. (Continuation)

Case	Year	Age	Sex	Dgn	Segment	Method	Ach	Shortening (cm)	Lenghtening (cm)	+/–	Lenghtening (%)	Index	Response
30.	1985	4	m	5	2	2		1	3	2	12.5	35.7	+
31.	1985	14	f	2	1	3		4	5	1	11.9	39.4	
32.	1985	20	f	5	1	3		6	6	0	16.2	25.5	+
33.	1985	17	f	3	1	2		9	9	0	25.7	53.6	
34.	1986	11	f	2	2	2		5	6	1	19.4	13.5	
35.*	1986	13	f	1	2	2			5	5	22.7	20.0	
36.	1986	16	m	2	1	2		7	6	–1	12.8	38.8	
37.	1986	8	m	2	1	2		6	6	0	12.8	38.8	
38.	1986	4	f	2	1	2		6	5	–1	20.0	40.8	
39.	1986	13	m	3	1	2		5	5	0	11.2	36.2	+
40.	1987	7	m	4	1	2		5	6	1	30.0	29.3	+
41.	1988	14	f	2	1	3		5	5	0	11.9	33.4	+
42.*	1990	8	m	1	2	2			11	11	68.7	15.1	+
43.*	1990	9	f	1	2	2			11	11	73.3	16.1	+
44.*	1991	5	m	2	2	3		7	10	3*****	39.8	47.2	+
45.	1991	8	f	2	1	2		4	4	0	11.4	23.5	
46.	1993	16	f	2	2	2		8	8	0	20.5	27.4	+
47.	1994	14	f	4	2	2		4	4	0	10.5	28.8	+
48.	1994	18	f	2	1	3		4	4	0	8.5	31.8	+
49.	1995	24	f	2	1	2		6	6	0	13.1	28.2	+
50.	1995	17	f	2	2	3		7	7	0	15.6	59.4	+
51.	1995	8	f	5	1	3		4	4	0	11.4	34.5	+
52.	1995	9	m	2	2	3		5	5	0	12.5	20.0	+
53.	1995	8	m	2	2	3		7	7	0	22.0	27.0	+

Dgn=diagnosis; 1–achondroplasia; 2–congenital; 3–other (M.Ollier's, OI, SFCE, paralysis); 4–sequelae of osteomyelitis; 5–posttraumatic shortening. Segment=lengthened limb segment; 1–femur; 2–tibia. Method=method of operation; 1–distraction epiphysiolysis; 2–metaphyseal corticotomy; 3–metaphyseal osteotomy. Tendo=tibial lengthening with lengthening of the Achilles tendon. +/– over or under-lengthening. Response=patients who responded to the questionnaire. * – limb lengthening in both legs in achondroplasia cases; ** – patients undergoing two lengthenings, *** – patient undergoing three lengthenings, each 5 cm; ****– lengthening percentages excluded from the calculation of mean value because the main reason for the procedure was deformity correction but not limb lengthening; *****– patients undergoing overlengthening to compensate for the difference in length in the period of rapid growth; at the end of surgery the limbs were equal.

For detection of metaphysis, the AO/ASIF quadrate method (Müller *et al* 1987) was used. In cases of corticotomy the bone was not broken, to prevent damage of the cancellous bone and the vessels inside it. For this purpose a special crescent-shaped osteotome was designed (Invention No1297826 USSR) consisting of a handle and a cutting edge covered with a plate which protects soft tissues and prevents incision of the spongious bone. The bone is detected using a longitudinal periosteal incision. The cutting edge of the instrument is placed onto the bone. Transversal corticotomy is performed with hammer blows on the handle of the osteotome below the periosteum (Fig 2).



A B

Figure 2. A – osteotome; B – performing of corticotomy

Distraction began on the third to the fifth day at a rate of 1 mm per day in four equal doses applied. Distraction rate was decreased if there was radiographic evidence of poor bone formation, or if the patient complained of excessive pain.

Follow-up visits were scheduled for every week during the distraction period and monthly during the consolidation phase. Monthly x-rays were taken for assessment of the ossification of the regenerate. The last x-rays were taken at 2 years of follow-up, and also later if the patients had complaints. Modified criteria for analysing the distractional gap, as described by Fischenko and the coauthors (1976), were used. The estimated six stages of bone tissue maturity were: I-soft tissue stage, IIA-initial calcification of the regenerate, IIB-final calcification, IIIA-initial ossification, IIIB-final ossification, IV-remodelling of the bone tissue.

Complications were classified in the simplest way into minor and major and were registered. Minor complications were transient swelling and pin surrounding infection; major complications required additional surgical treatment or affected the final result (pseudoarthrosis, delayed regeneration, fracture or deformity of the regenerate, ankylosis or subluxation of the joint and osteomyelitis). Transient restriction of the movement of the joints were not regarded as a complication but as a sign of limb lengthening.

Physical treatment and dynamization was applied to every patient, partial to full weight bearing was allowed as tolerated with the external fixator in place.

The percentage of lengthening (length of the distraction gap divided by the prelengthening length of the bone segment multiplied by 100%) and lengthening indices (days needed for lengthening/centimetres of lengthening) were calculated.

After fixator removal, most limbs were further protected for four to six weeks in an external orthosis or cast and physiotherapy was applied.

4.2.2. Elecrtromyographic study

4.2.2.1. Patient data

Seven patients who had had a distraction lengthening of the femur participated in the study after having given informed consent. Before and after lengthening, the lengths of the lower extremities had been measured as the distance between the anterior superior iliac spine and the medial malleolus at the ankle; the average percentage of lengthening (and standard deviation) had been 16 ± 4.7 per cent (range 11 to 24 per cent) (Table 4). Limb length discrepancy was congenital in five patients (cases 1 through 5) and acquired in two (cases 6 and 7). Three patients (cases 1, 2, and 3) had had lengthening with the Ilizarov technique (Ilizarov and Trohova 1973, Paley 1988) and four (cases 4 through 7), with the Wasserstein method (1971; 1990). Immediately after the lengthening, the knee joint had been stiff in all of the patients, but a full range of motion of the joint was restored soon after the procedure. No other major complications or symptoms of peripheral nerve injury had been observed during the limb lengthening. All patients had completed postoperative rehabilitation programme.

Table 4. Data of the patients of the electromyographic study

Case	Age (yrs)	Year	Preop discrepancy	Percentage of lengthening	Distraction rate (cm/day)	Duration of distraction (days)	Atrophy index (percentage)	Time to full weight bearing (wks)
1	14	1985	4	12	0.0253	158	1.9	52
2	8	1984	5	20	0.0370	135	3.8	29
3	16	1986	7	13	0.0257	272	4.2	26
4	18	1981	7	15	0.0337	208	3.1	36
5	28	1982	5	11	0.0495	101	2.4	32
6	16	1982	9	24	0.0441	204	5.8	32
7	21	1978	7	17	0.0901	78	3.9	52

Physical examination of the patients, performed before electromyographic testing, showed that all of the knees had a range of motion of 0 to at least 120 degrees of flexion. There were no symptoms of peripheral nerve injury. Although all of the patients considered their condition good, all described a feeling of discomfort in the knee or hip of the involved limb when they were overly fatigued.

To quantify the possible atrophy of the muscles of the thigh on the side of the lengthening, the circumference of the thigh was measured five, ten, and fifteen centimeters proximal to the lateral epicondyle of the femur. These measurements were added up and an atrophy index, expressed in percentage, was calculated as 100 – (100 x (the sum of the three measurements of the operated thigh/the sum of the three measurements on the nonoperated thigh)).

It was assumed that the muscles on the nonoperated side were of normal size.

4.2.2.2. Electromyographic testing and analysis of the data

The patients sat in a specially designed chair with a backrest that was inclined 20 degrees from the vertical and a seat that was inclined 20 degrees from the horizontal so that the hip was flexed 90 degrees. Two straps, one that secured the pelvis and the other that secured the upper part of the body, were used to keep the angle of the hip joint constant. A pad was placed anteriorly to the inferior third of the thigh to prevent motion proximal to the knee. A strap was placed around the leg so that a load could be applied through the strap to a point just proximal to the malleoli of the ankle; this load was 15 per cent of the body weight of the patient and ranged from eight to twelve kilograms. The patients had acquainted themselves with the procedure before testing. For the test, the knee was actively extended by isometric contraction of the knee extensors and kept at an angle of 30 degrees of flexion, with the load applied, for thirty seconds. A visual feedback system ensured that this angle did not deviate by more than 5 per cent during the test. The involved and normal sides were tested in a random order.

Pairs of silver-silver chloride electromyographic surface electrodes (type Q-00-R; Medicotest, Olstykke, Denmark) were applied over the most prominent bellies of the vastus medialis, rectus femoris, and vastus lateralis muscles. The centre-to-centre distance of an electrode pair was 40 millimeters. A common-ground electrode was placed apart in an electrically indifferent area. The electromyographic signals were recorded with a Muscle Tester ME 3000P microprocessor (Mega Electronics, Kuopio, Finland). The raw electromyographic signal was amplified, full-wave rectified, and smoothed with a low-pass filter at a bandwidth of ten to 1000 Hertz before being stored for additional analysis.

Electromyographic data were analysed with a specially designed computer software (Muscle Tester Analyze Software, version 1.20; Mega Electronics).

Raw electromyograms were checked with respect to so-called cross talk before analysis; a random change of phase between the electromyograms of a muscle pair ensured the absence of cross talk.

The myoelectric power density spectrum as well as its simple estimate, median frequency, were calculated with the fast Fourier transformation, as described by Hagg (1991), for the entire record as well as separately for the first three-second segment. The increased median frequency of this segment was evaluated as an index (percentage) of motor-unit recruitment. A number of investigators (Lindstrom *et al* 1970; Stulen and DeLuca 1981; Solomonow *et al* 1990) have suggested the use of median frequency as an index of motor-unit recruitment because of its dependence on the conduction velocity of the motor units. Since the size of motor units may vary among the knee extensor muscles, as implied by their different fibre distributions (Johnson *et al* 1973), average conduction velocity and, consequently, the range of the change in median frequency may also vary. Therefore, the change in median frequency was normalized with respect to the peak median frequency of a particular recording to avoid misinterpretation when the rates of the change in the median frequency of different muscles were compared. Peak median frequency was established as the highest value of the median frequency of a half-second period. Any shift in the myoelectric power density spectrum of the entire record to lower frequencies (decreased median frequency, expressed in units per minute) was considered as a sign of localized muscle fatigue, in accordance with earlier studies (Stulen and DeLuca 1981; Bigland-Ritchie *et al* 1986; Hagg 1991).

4.2.3. Follow-up questionnaire

A questionnaire developed originally by Tjernström and Rehnberg (1994) modified for this specific study was used.

Follow-up questionnaire				
I Case				
Age				
Sex				
employed	studying	unemployed	disabled	home bound
II Complaints				
1. Back pain:				
<u>Pain</u>		<u>Appearance</u>		<u>Localization</u>
absent		none of the time		thoracic
a little		a little of the time		lumbar
moderate		some of the time		total
serious		most of the time		
extreme		all of the time		

2. Hip pain:		
Pain absent a little moderate serious extreme	Appearance none of the time a little of the time some of the time most of the time all of the time	Localization right hip left hip both
3. Knee pain:		
Pain absent a little moderate serious extreme	Appearance none of the time a little of the time some of the time most of the time all of the time	Localization right knee left knee both
4. Limping	Appearance	
	1. none of the time	
	2. a little of the time	
	3. some of the time	
	4. most of the time	
	5. all of the time	
III		
1. Do you remember what cumbered you before limb lengthening?		
2. Do you remember how you compensated for your limb length? (scoliosis, pelvic tilt, flexed knee, equinus, heel lift, supporting brace)		
3. Did you have pain before limb lengthening?		
4. Did you have pain during limb lengthening?		
5. Are you satisfied with the present situation?		
	1. Fully satisfied	
	2. Partly satisfied	
	3. Not at all satisfied	
6. Are you satisfied with the present cosmetic situation?		
	1. Fully satisfied	
	2. Partly satisfied	
	3. Not at all satisfied	
7. Do you wear a heel lift?	Yes	No
8. If you needed to pass the limb lengthening once more, would you agree?		
	1. Yes	
	2. No Why?	

4.3 Statistical analysis

4.3.1. Statistical analysis of the histological samples

Statistical analysis was performed using mainly the unpaired t-test at the level of significance p less than 0.05 ($p<0.05$). In some cases (comparison between the groups, etc.) ANOVA with the Newman-Keuls multiple comparisons test was applied.

4.3.2. Statistical analysis of the electromyographic studies

Differences in the mean values were analysed with the Student t-test for paired data (for the contralateral muscles) and for independent data (for the unilateral muscles). The Wilcoxon signed-rank test was applied to evaluate individual differences in muscle fatigability. Correlation analysis was performed to assess the relationship between the variables. In all of the statistical analyses, the level of significance (alpha) was considered to be 0.05. As the study involved 53 comparisons and the minimum level of significance was 0.05, there was 1 – (1 – 0.05)53, or 93 per cent chance for at least one incorrect result of statistical analysis experimentwise.

4.3.3. Statistical analysis of the questionnaire data

Where it was appropriate, t-test was employed for paired or unpaired data.
In order to obtain clear and comparable data about pain and subjective satisfaction on the basis of the follow-up questionnaire, the following scoring was used:

1) pain intensity was scored so that “absent“ was marked as “1“ and “extreme”, as “5“.
2) appearance of pain was scored so that “none of the time“ was marked as “1“ and “all of the time”, as “5“.
3) satisfaction was scored so that “fully satisfied“ was marked as “1” and “not at all satisfied“, as “3”.
4) limping was scored so that “none of the time“ was marked as “1” and “all of the time“, as “5”.

These scores were used in calculations with continuous data and in t-statistics for the patient groups. Depending on group size and number of divisions, Fisher’s exact test or the chi-square test was applied. With all the methods a p-value less than 0.05 was considered statistically significant. Regression analysis was performed for comparison between the groups to assess the affecting factors.

5. RESULTS

5.1. Experimental study

5.1.1. Distraction bone healing after corticotomy (rabbit model) (publ I)

Experimental bone regeneration after corticotomy of the distal metaphysis of the tibia and gradual lengthening at a daily rate of 0.35 mm was analysed in 20 rabbits. Maximum lengthening of the tibia was 10 mm. The control group constituted investigation of the tibia on the opposite (normal) side. Histological studies of the regenerate have shown variability in the size of bone trabeculas surrounded with osteoblasts. Bone regeneration is characterized by the process of osteoclastic resorption. The quantity of osteoclasts surrounding the bone trabeculas of the regenerate was estimated. According to the data, there were 60.3 ± 0.9 osteoclasts immediately (first 5 days) after the operation compared with 24.2 ±5.7 osteoclasts in the control group. After 7 days the amount of osteoclasts diminished nearly to that in the control group with a new rise in their amount after 10–14 days. A normal quantity of osteoclasts was reached after 30 – 35 days (Fig. 3).

Increase in osteoclasts after 5, 10, 14 and 21 days of the experiment was proved by statistical analysis ($p < 0.01 – 0.05$). Hence, bone regeneration is characterized by proliferation and destruction processes, while regeneration of the lengthened tibia was more rapid compared with the tibia on the normal side. The experimental data showed that there were 71.7 ±5.8 % of bone trabeculas from all tissues on the side of bone lengthening compared with 46.9 ±6.5 % on the control (normal) side. These data confirm the opinion that nonoperated bone does not show such high level of bone ossification activity that is typical of operated bone. Continuous irritation of operated bone stimulates aseptic inflammatory reaction followed by intensive differentiation of various tissues including the bone tissue. Histological investigations showed that venous hyperaemia took place during the first week after operation. This indicates that method of the operation used did not cause disorders of circulation.

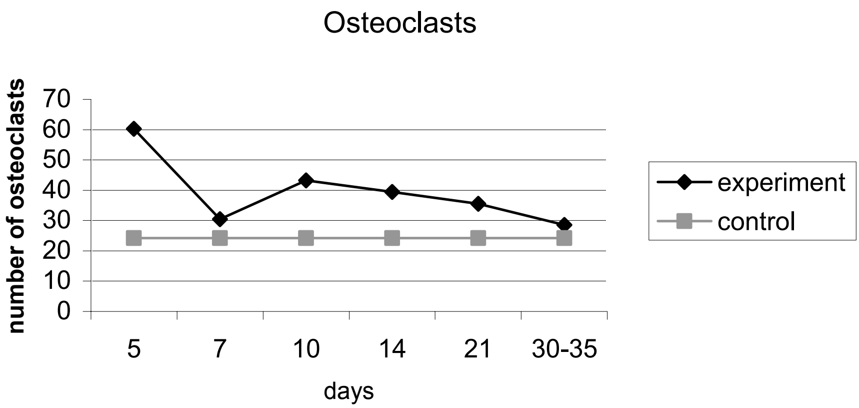


Figure 3. Number of osteoclasts around newly formed bone

5.1.2. Bone healing after injury (rat model) (publ II)

Filling of 18 resected tibial defects with a length of 4mm is most similar to embryohistogenesis with periosteal primary and endosteal secondary ossification. Removal of a bone fragment caused local hemorrhage and tissue destruction with simultaneous early neoangiogenesis. Reparative changes also occurred in soft tissues. The granulation tissue became gradually denser and fibroreticular (4th – 7th day), on the basis of which the chondrous tissue was formed. This newly formed metaplastic tissue was referred to as the periosteal and endosteal fibrous callus(7th day) and later as the chondrofibrous callus (14th – 21st day). The latter (28th day) forms a spindle-shaped thickening surrounding osteotomy and filling also the gap between the two ends of tibia fragments – the peri- and endosteal young bone callus, respectively (Fig. 4). The definitive spongy bone callus was formed later (35th – 42nd day), after partial replacement of the chondrofibrous and chondrous callus.

In the group of bone cortex perforation with a diametre of 1.5 mm in the intact periost in 25 animals endosteal osteogenesis was primary and periosteal osteogenesis was secondary.

Perforation of the bone caused, like in the other groups, hemorrhage and tissue destruction on the 1st – 4th day but reparative changes already on the 4th – 7th day. This occurred in soft endosteal tissues. Fibroblasts began to intensively synthesize extracellular substance including collagen. On the 14th day collagen synthesis was visible and intensive in osteoblasts (packed collagen fibrils in vacuolated cytoplasm). Posttraumatic osteohistogenesis was completed by the 21st day, but organomorphogenesis (bone formation) and remodelling continued up to the 28th – 42nd day. Composition of bone on the 28th day of the experiment was already over 84%. Bone repair after perforation had one peculiarity: endosteal ossification followed directly without the chondrous stage, whereas periosteal ossification occurred through the chondrous callus (Fig. 5). The growth and differentiation of the endosteal callus was inhibited and the periosteal callus was mostly arrested in the groups with the isolated periost (Figs. 6; 7).

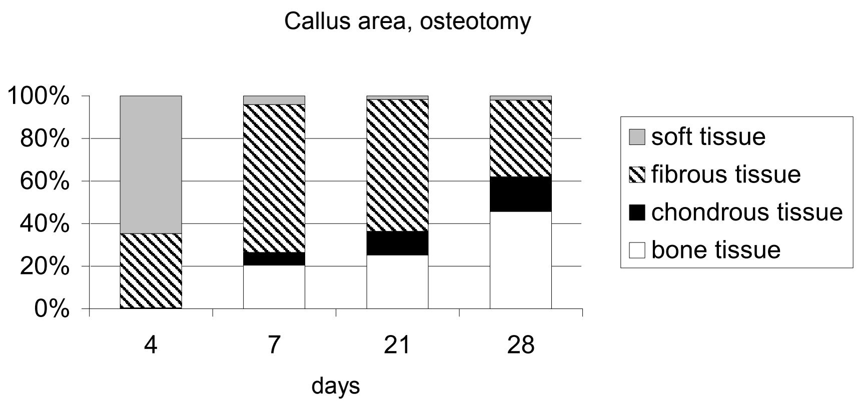


Figure 4. The area of callus tissues 4–28 days after osteotomy in rat tibia (percentage of the total callus area)

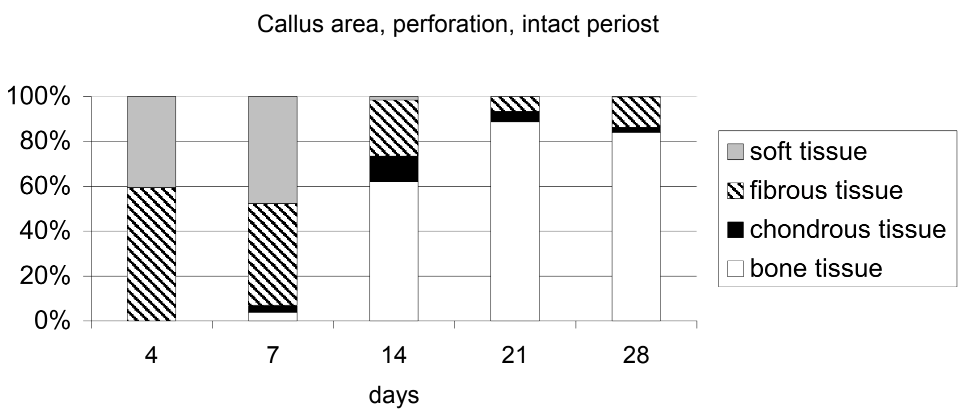


Figure 5. The area of endosteal callus tissues 4–28 days after perforation with the intact periost (percentage from the total callus area)

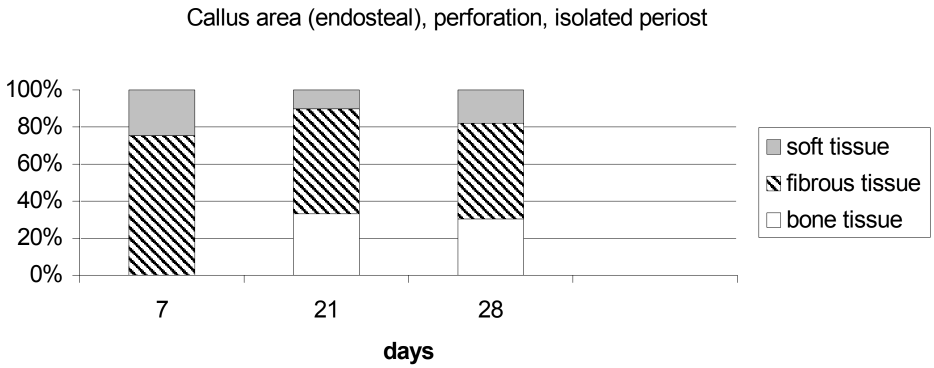


Figure 6. The area of endosteal callus tissues 7–28 days after perforation with isolation of the periost (percentage from the total callus area)

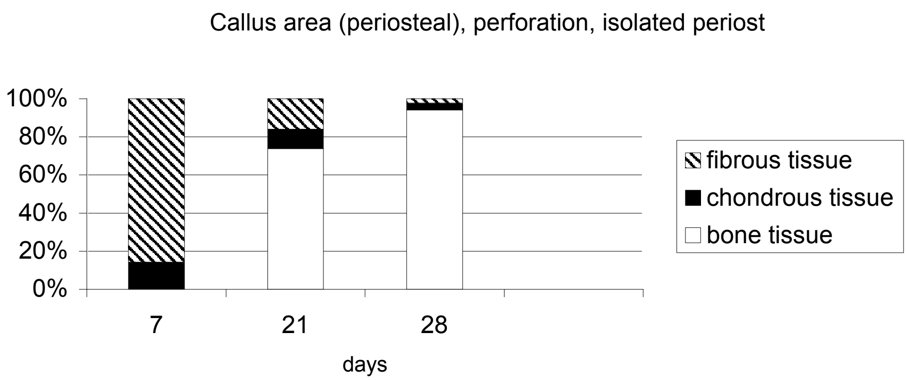


Figure 7. The area of periosteal callus tissues 7–28 days after perforation with isolation of the periost (percentage from the total callus area)

5.2. Clinical assessment of lower limb lengthening

5.2.1. Electromyographic assessment of limb lengthening (publ III)

Surface electromyography of the quadriceps femoris muscle was performed. The lengthening effect of the extensors of the knee on the femur was the following. Average increase in the median frequency of the knee extensor muscles of the lengthened thigh was significantly smaller ($p < 0.001$ for each comparison) during the first three seconds of the test compared with that on the normal side (Fig. 8). The vastus medialis exhibited a significantly smaller average increase in median frequency ($p < 0.01$) than the rectus femoris and the vastus lateralis on the lengthened side. No significant differences were observed in peak median frequency between the muscles on the normal side. The extensor muscles on the lengthened side showed significantly higher fatigability ($p < 0.01$ to 0.05) than those on the normal side (Fig. 9). The vastus medialis was significantly more fatigued ($p < 0.05$) than the rectus femoris and the vastus lateralis on the lengthened side, but no significant differences were observed between muscle fatigabilities on the normal side.

Analysis revealed excellent correlation between the percentage of bone lengthening and the fatigability of the knee extensors on the lengthened side (r (correlation coefficient) = 0.92 to 0.70; $p < 0.01$ to 0.05) as well as between preoperative limb length discrepancy and the fatigability of the lengthened muscles ($r = 0.85$ to 0.68; $p < 0.01$ to 0.05). The correlation was moderate between the percentage of bone lengthening and the motor unit recruitment of the muscles on the lengthened side ($r = -0.67$ to -0.64; $p < 0.05$ to 0.06). The atrophy index correlated well with the data for fatigue ($r = 0.88$ to 0.67; $p < 0.01$ to 0.05), preoperative limb length discrepancy ($r = 0.96$; $p < 0.001$), percentage of bone lengthening ($r = 0.87$; $p < 0.01$), and motor unit recruitment of the muscles on the lengthened side ($r = -0.84$ to -0.69; $p < 0.01$ to 0.05).

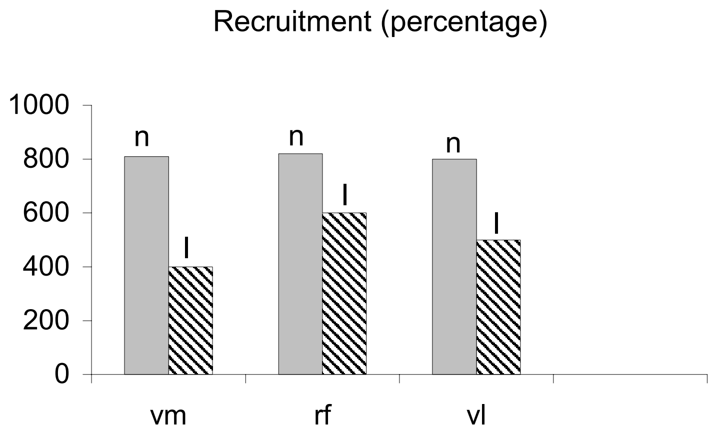


Figure 8. Graph of the mean percentage of motor unit recruitment in the knee extensor muscles of seven patients after unilateral femoral lengthening. The difference was significant between the muscles on the lengthened (l) side and those on the normal (n) side ($p < 0.001$ for each comparison) and between the vastus medialis (vm) and the vastus lateralis (vl) as well as between the vastus medialis and the rectus femoris (rf) on the lengthened side ($p < 0.01$ for both comparisons). vm-n = vastus medialis, normal side; vm-l = vastus medialis, lengthened side; rf-n = rectus femoris, normal side; rf-l = rectus femoris, lengthened side; vl-n = vastus lateralis, normal side; and vl-l = vastus lateralis, lengthened side

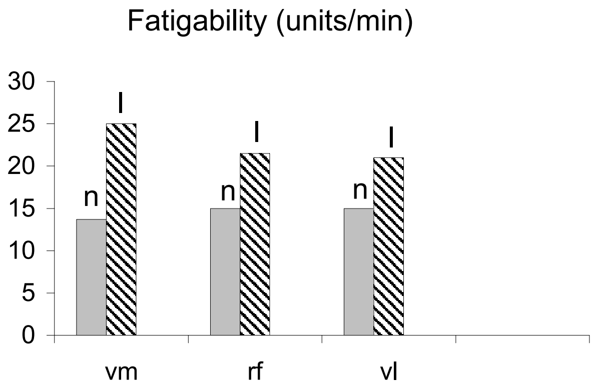


Figure 9. Graph of the mean fatigability (in units per minute) of the knee extensor muscles of seven patients after unilateral femoral lengthening. The difference was significant between the muscles on the lengthened side and those on the normal side (vastus medialis (vm), $p < 0.01$; rectus femoris (rf), $p < 0.05$; and vastus lateralis (vl), $p < 0.05$), between the vastus medialis and the rectus femoris on the involved side ($p < 0.05$), and between the vastus medialis and the vastus lateralis on the lengthened side ($p < 0.05$). vm-n = vastus medialis, normal side; vm-l = vastus medialis, lengthened side; rf-n = rectus femoris, normal side; rf-l = rectus femoris, lengthened side; vl-n = vastus lateralis, normal side; and vl-l = vastus lateralis, lengthened side

5.2.2. Limb lengthening in posttraumatic cases (publ IV)

There were 14 patients aged between 4–18 years who needed the lower limb lengthening. The patients were divided into four different groups:

Group I (4 patients) – minor lengthening (1–3 cm) with a correction of angular deformity (mean ~20°). Usually a plaster cast was applied for 1–2 months. The mean lengthening index was 29.9 d/cm (days per centimetre).

Group II (3 patients) – bone transport. All defects were infected. In one case (28 cm) tibial double level leg lengthening was performed. In this case 14.4 d/cm lengthening index was gained, but afterwards the patient needed a plaster and walking cast for 180 days. The lengthening index was 21.7 d/cm. No complications were observed.

Group III (3 patients) – femoral lengthening. The third corticotomy in the third time lengthened patient failed and procedure was taken as osteotomy. Callus formation was first seen at 4 weeks after corticotomy. The lengthening index for the whole group was 33.5 d/cm, with an exception of the last lengthening (46.4 d/cm) in the patient whose leg had been lengthened three times. Otherwise the lengthening index would have been 26.3 d/cm. Three times operated patient had minor complication.

Group IV (4 patients) – lengthening by using a tubular bone allograft (Wasserstein's procedure). At the end of the distraction calcification of the callus initially formed from the periosteum, a well-formed tubular regenerate was observed roentgenologically. After the second operation, a fusion of the allograft from the ends and gradual incorporation from the side of the periosteal tube appeared. Three of these patients had complications, two of them serious: persistent stiffness of the knee joint and necrosis of the wound after bone grafting. The lengthening index was 20.5 d/cm. All these patients had had a plaster cast for at least 60 days and a walking cast afterwards.

Taking into account all groups, complication rate was 28%: 14% accounting for “minor” complications as pin surrounding infection and 14% accounting for “major” complications as stiffness, wound necrosis. None of the complications affected the final result. One patient had an operative release of the knee joint. The other patients improved the movement of the joints. All patients were satisfied with the final result despite having complications.

5.3. Long-term follow-up

5.3.1. Long-term results of extensive limb lengthening in Ollier’s disease (publ V)

Extensive lower limb lengthening was performed in a 14-year-old male patient with the diagnosis of Ollier’s disease. The anatomical shortening of the right lower limb constituted 32 cm − 22 cm of the thigh and 10 cm of the leg. A varus deformity of the femur and a valgus deformity of the tibia were evident. The extension-flexion range of the knee joint was 0°/0°/90°. Radiological investigation showed focal areas of the cartilage in the metaphyseal parts of the long bones and in the pelvis. Histological investigation of the femur showed evidence of embryonic cartilage cells. The patient used a limb orthosis.

Metaphyseal distraction was started after distal femoral corticotomy, performed by using a special chisel through Ollier’s focus. After eight months, a 22 cm lengthening of the femur (75% of initial length) was achieved. Radiologically, a good bone regenerate was formed. No evidence of vascular or neural disturbance was found. A two-stage lengthening of the tibia by Wasserstein was followed. The external fixator remained unremoved on the thigh. Distraction rate was 1.5 mm daily. After 2 months, a 10 cm lengthening of the leg was achieved. In the second stage the bone defect was filled with a cylindrical bone allograft. The distraction devices were removed both from the thigh and from the leg in 1.5 years after the beginning of femur distraction. The lengthening indices were calculated being 22.5 days per centimetre for the femur and 21 days per centimetre for the tibia (altogether 15.5 days per centimetre). Fine needle biopsy investigation showed that most embryonic cartilage cells had been replaced with the bone tissue. Five-year follow-up showed a good functional result. The range of knee extension-flexion was 0°/0°/60°. Follow-up after 25 years (in 2004) confirmed the same functional result but with a painful knee. The host bone had incorporated the allograft. Signs of osteoarthritis were observed on a knee radiograph.

5.3.2. Long-term results and patient satisfaction in limb lengthening (publ VI)

A database was created containing the stored follow-up data of the patients and the questionnaire data. Altogether 63 lower limbs were lengthened in 53 patients (total 72 operations). As the objective indicators (L%, LI, ROM, range of complications, gender/sex, type of operations, etiology) were the same as one to two years after limb lengthening, they were compared here with the data of long-term satisfaction.

The amount of lengthening of one limb segment was 1–22 cm. In minor cases mostly angular deformity correction was done. There were altogether 26 complications (in 49% of patients), among which 15 (28%) were minor in and 11 (21%) were major complications.

A mean 26.2% (range 8.5–81) of lower limb lengthening was gained. The percentage of limb lengthening differed significantly in achondroplasia cases, 65.3±7.9%, versus congenital cases, 16.0±0.8% (t-test; $p<0.0001$) and versus sequelae of osteomyelitis cases, 20.2±2.8% (t-test; $p=0.0029$). The mean lengthening index was 29.5 (range 12.9–59.4) days per centimetre but it varied between the study groups. The lengthening indices differed between the diagnosis groups as well. A significant difference in the lengthening indices was found between the groups of achondroplasia (22.2±2.0 d/cm) and congenital shortening (33.6±2.3 d/cm) (t-test; $p=0.0088$) and between the group of congenital shortening and sequelae of osteomyelitis (23.7±1.9) (t-test; $p=0.0245$) (Table 5).

Table 5. Lengthening percentage and lengthening index depending on diagnosis.

Diagnosis (number of patients)	Lengthening percentage	Lengthening index (days per centimetre)
Achondroplasia (6)	65.3±7.9	22.2±2.0
Congenital shortening (27)	16.0±0.8	33.6±2.3
Sequelae of osteomyelitis (8)	20.2±2.8	23.7±1.9
Posttraumatic shortening (8)	13.5±1.1	30.4±2.4
Others (4)	32.6±14.7	33.4±7.5

A questionnaire was sent to 53 patients. Response rate was 70% (37 patients); 15 men and 22 women responded. Four patients with achondroplasia, 17 with congenital shortening, 6 with sequelae of osteomyelitis, 7 with posttraumatic shortening and 3 patients with other diagnoses responded.

Mean follow-up time was 19.4 years (range 10–29). At present only one of the examined patients has disability, three of them (all women) are at home nursing children and the rest are employed or attend school. The majority, 26 patients (70%), remember that something cumbered them before the surgery. The reasons for cumbering were: difference in the length of the lower legs in 15 patients, limping in 5 patients, short stature in 3 patients and other in 3 patients. For compensation for the deformity, more than half of the patients, 54% (20 patients), used walking aids or a shoe lift. Despite the fact that 24 (65%) of the respondents are limping most of the time or all of the time, 29 (78%) are satisfied with the present situation and 25 (68%) are satisfied with the cosmetic appearance. If there arose the need to pass limb lengthening once more, 30 (81%) of the examined patients would agree to do it. Although 81% of the patients agreed to pass limb lengthening once more if were needed, this decision showed no significant difference with respect to any objectively estimated functional indicator one year postoperatively (Table 6).

After limb lengthening the function of the knee joint was impaired as expressed by the diminishing of knee flexion by a mean of 79.1º±5.3º, which differed significantly from the preoperative values (t-test; $p<0.0001$). The degree of the impaired function of the knee did not significantly differ for the method of operation used. After one year the function was significantly improved, but flexion was still 12.3º±4.0º less than the preoperative values (t-test; $p=0.0103$). The patients in the group of distraction epiphysiolysis gained almost the same level of knee flexion after one year, 143º±2.1º, versus preoperative 145º±1.7º (t-test; $p=0.47$). The patients with metaphyseal osteotomy showed a larger difference between the preoperative (128.2º±7.6º) and one-year postoperative (115.0º±7.6º) knee flexion values, however, this difference did not reach statistical significance (t-test; $p=0.23$). A statistically significant difference in knee flexion was found in the metaphyseal corticotomy patients: the preoperative value being 136.5º±2.6º and the one-year postoperative value being 121.7º±5.6º ($p=0.0186$).

Mainly low back pain was observed in 15 patients, but it was considered serious only in two patients. Hip pain was noted in 11 patients and it was serious or extreme in 2 cases. Knee pain was observed in 18 patients and it was serious or extreme in 3 cases. At the same time, 13 patients stated that they did not have any pain in any segment of the limb or in the back. Among them 2 patients had achondroplasia, 4 had congenital shortening, 2 had sequelae of osteomyelitis and 4 had posttraumatic shortening.

The score of the intensity of back pain for the patients who underwent distraction epiphysiolysis was 2.50±0.62 (on the border between "a little" and "moderate"), 1.40±0.15 for the corticotomy patients (on the border between “absent” and “a little”) and 1.73±0.30 for the osteotomy patients (on the border between “absent” and “a little”). A statistically significant difference was detected between the distraction epiphysiolysis group and the corticotomy group (t-test; p=0.0168).

Satisfaction with the present situation differed in the context of the method of operation. The satisfaction scores were 2.33±0.33 in distraction epiphysiolysis, 1.7±0.18 in metaphyseal corticotomy and 1.18±0.18 in metaphyseal osteotomy. This indicates that patients who had undergone metaphyseal osteotomy and corticotomy were more satisfied. The difference in satisfaction between the groups of metaphyseal osteotomy and distraction epiphysiolysis was statistically significant (t-test; p=0.0046) (Fig.10).

Difference in satisfaction with the present situation between the genders was absent. Twenty-five patients (13 men; 12 women) who were generally satisfied with the cosmetic result, dissatisfied 12 (2 men; 10 women). This difference was statistically significant (Fisher’s exact test, p=0.043) (Table 6). Fisher’s exact test for the appearance of disturbing scars showed a statistically significant difference between the genders (p=0.029). Nineteen patients (11 men; 8 women) were satisfied and 18 patients (4 men; 14 women) were dissatisfied with the appearance of scars.

Comparison of 22 (21 satisfied; 1 nonsatisfied) patients who had no complications and 15 (8 satisfied; 7 nonsatisfied) patients who had complications revealed a statistically significant difference (Fisher’s exact test; p=0.004). Hence the patients without complications were more satisfied with the present situation. A similar significant correlation was found between complications and satisfaction with the cosmetic effect (Fisher’s exact test; p=0.030). Patients who did not limp (13) were all satisfied with the present situation in comparison with the 24 patients who limped among whom 8 were dissatisfied with the present situation (Fisher's exact test; p=0.019)

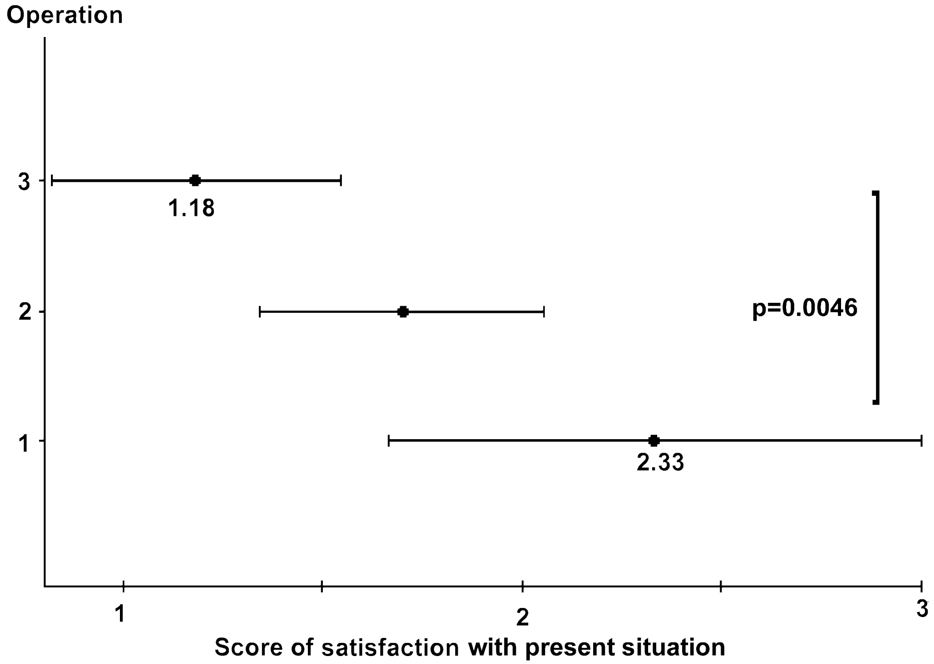


Figure 10. Score of satisfaction with present situation. 1 – distraction epiphysiolysis; 2 – metaphyseal corticotomy; 3 – metaphyseal osteotomy. Bars indicate standard deviation from the mean score.

Among the 17 patients who had used a heel lift or a brace and had had some compensative deformity before limb lengthening, only one would not have agreed to pass limb lengthening operation once more. Among the 20 patients who did not use any aid before the surgery, six would not have agreed to pass the surgery once more (chi square test; p=0.035).

For the patients who would agree to pass limb lengthening once more, the score of hip pain in the present study was 1.37±0.15 (on the border between "absent" and "a little") and for those who would not agree to undergo this procedure once more, the corresponding score was 2.72±0.52 (on the border between "a little" and "moderate") (t-test; p=0.0015). A similar difference was observed in the score of appearance of hip pain between those who would agree (1.47±0.18; on the border between "none of the time" and "a little of the time") and those who would not agree to pass the surgery once more (2.43±0.37; on the border between "a little of the time" and "some of the time") (t-test; p=0.028). At the same time, it should be mentioned that neither pain intensity nor appearance of pain in the hip affected satisfaction with the present situation.

Table 6. Data of the patients who responded to the questionnaire; Fisher's exact test applied to data analysis; only p<0.05 values are marked in Table.

		Satisfaction with present situation		Satisfaction with cosmetic effect		Readiness to pass LL once more	
		Satisfied/ Non satisfied	p-value	Yes/No	p-value	Yes/No	p-value
Sex	Male	13/2	0.431	13/2	0.043	13/2	0.677
	Female	16/6		12/10		17/5	
Compli-cations	Yes	8/7	0.004	7/8	0.036	11/4	0.408
	No	21/1		18/4		19/3	
Type of operation	DE	2/3		4/1		3/2	
	MO	13/2		11/4		13/2	
	MC	14/3		10/7		14/3	
Etiology	Acondroplasia	4/0		3/1		3/1	
	Congenital	12/5		10/7		11/6	
	Osteomyelitis	4/2		4/2		6/0	
	Posttraumatic	7/0		5/2		7/0	
	Other	2/1		3/3		3/0	
Limping	Yes	16/8	0.032	15/9	0.476	20/4	0.678
	No	13/0		10/3		10/3	

DE–distraction epiphysiolysis; MO–metaphyseal osteotomy; MC–metaphyseal corticotomy

6. DISCUSSION

6.1. Experimental study

The purpose of the present experimental study was to investigate the peculiarities of bone regeneration and the cellular composition of the regenerate during bone distraction lengthening. Also the response of the cortical bone, periost and endost to resection osteotomy or cortical perforation was in the focus of this study, which serves as a model of distraction bone lengthening both in experimental and clinical research.

Experiments were carried out on rabbits and rats. The choice of rats and rabbits as the experimental animals in this study was reasonable. Several studies have demonstrated that the histological pattern of bone formation by distraction osteogenesis in dogs, rabbits, rats, and mice is analogous to that in humans (Aronson 1994; Tay *et al* 1998; Aronson *et al* 2001),

Experimental corticotomies carried out on the rabbit tibia in the present study demonstrate early endosteal regeneration where osteoclasts appeared already on the 5th day. Chambers (1980) also established that in the case of activated osteogenesis the number of osteoclasts on bone trabeculas increased. On the 14th day of distraction, activated osteoclasts, as well as an increased number of small blood vessels, were detected in the distraction gap (Ilizarov and Berko 1980). In another investigation of lengthening of the tibia, a rise in the amount of osteoclasts was seen only on the 35th day of distraction (Ilizarov *et al* 1975). In corticotomy, early bone regeneration and osteclastic activity are the result of osteoblastic positive feedback. Multinucleated cells were considered osteoclasts as their differentiation stage was not investigated. Early-appearing large multinucleated cells can be nondifferentiated giant cells and the appearance of osteoclasts is noted only at the end of the first week (Landry *et al* 1996).

In the present studies, no pseudoarthrosis associated with the stability of the minifixator was observed. The stability of the distractor is important (Ilizarov *et al* 1969). In stable distraction conditions bone regeneration is initiated from the endosteal side by the nondifferentiated cells of bone marrow (Ledyaev 1975).

In experiments with rabbits in the present study, venous hyperaemia was observed but it returned to normal, in comparison with the control, during the first week. This demonstrated undamaged circulation as well. The first reaction was mostly connected with operation trauma. Sveshnikov and the coauthors (1985) demonstrated the important role of circulation in bone regeneration. As this finding shows mostly reaction to operation, but not damage to blood circulation in the bone, it serves as a good basis for bone regeneration. Probably, there was no damage from the pins of the external fixator as they were introduced through the metaphysis in the experiments with rabbits. The nutritional arteries were not damaged by the pins, which may happen, according to an earlier study (Shugarov and Arapov 1975).

Posttraumatic bone repair, including destruction, inflammation, resorption, reparation and remodelling, is a normal wound healing process of mesenchymal tissues. It varies according to the character and degree of injury.

The means of choice for exploring bone regeneration in the present study was resection bone defect of rat tibia and perforation of tibia. Song and the coauthors (2002), when comparing the callus at a bone defect with the callus at the distraction site of simple lengthening, did not find a significant difference in bone quality in this case. In experimental filling defects Ilizarov and the coauthors (1975) observed that periosteal regeneration is more intensive compared to endosteal regeneration. However, in another investigation the same author found that endosteal regeneration takes place on the fifth day after diaphyseal osteotomy of the femur at the place of the bone cut (Ilizarov and Berko 1984). An important factor in bone repair is the intact periosteum (Landry *et al* 1996; Gordh *et al* 1997). Kojimoto and the coauthors (1988) showed that the periosteum is the most important structure in bone healing and intramembranous ossification will follow whether or not the medullary canal has been disrupted. Earlier Ilizarov and the coauthors (1969) demonstrated that bone formation would occur more rapidly and more securely if the endosteal structures were left intact.

Endosteal ossification was primary and periosteal ossification secondary after bone cortex perforation. The endosteal callus was intramembranous, whereas the periosteal callus was of the chondrous type. Isolation of the periost supressed periosteal osteogenetic activity and likewise the activity of the endosteal cells and osteoinductive cells of bone marrow as well as of the progenitor cells. Formation of the periosteal and the endosteal repairing bone callus was markedly inhibited appearing only on the 21st day, instead of the normal 4th–7th day.

According to the present study, it can be stated that in presence of bone marrow cells the best regeneration occurs from the endosteal and periosteal sides. The superiority of the periost and endost was discussed. This study revealed that the interaction of the periost and endost plays an important role. The similar findings have been reported by other investigators as well (Guichet *et al* 1998). Ilizarov and the coauthors (1984) found that the bone marrow cells are activated simultaneously with rapid bone regeneration, especially on the first 30 days of distraction. This is consistent with a study by Coleman and Scott (1991) who pointed out the importance of all structures in bone regeneration. In both experimental groups, like embryohistogenesis, repair processes have a similar histogenesis (fibrous, chondrous, bone tissue callus formation) and organogenesis (callus replacement, bone remodelling and recanalization). This statement has an important biological and clinical significance, besides its relevance in limb lengthening. The clinical limb lengthening methods used were developed on the basis of the experimental studies within this thesis.

6.2. Clinical study

Most limb lengthenings in the present study were performed in the metaphyseal region. For performing corticotomy we used a special curved osteotome invented by us; a similar instrument has been employed later by other researchers as well (Kristiansen and Steen 2002). Although in this study it was attempted to perform corticotomy in most cases, in some cases failure in operative technique or the fragility of bone did not allow to regard these cases as corticotomy but as osteotomy. Metaphyseal lengthening is preferred to diaphyseal lengthening since metaphyseal bone is expected to heal faster than diaphyseal bone (Fischgrund *et al* 1994). White and Kenwright (1991) concluded that the level of osteotomy is located at the junction between the metaphysis and the diaphysis, an area with a greater potential for bone healing than the mid-diaphysis. Stiffness of the knee joint in femoral lengthening was lesser when corticotomy was performed in the subtrochanteric region (Hankemeier *et al* 2004). According to another study, metaphyseal osteotomy was characterized by superior bone healing compared with diaphyseal osteotomy (Bowen *et al* 1993). Korzinek and the coauthors (1990) stated that when corticotomy at the metaphyseal level was generally successful, then in the diaphysis the same procedure did not result in a major disruption of intramedullary circulation in only 30% of cases.

In the present study the reason for starting lengthening on the 3rd or the 5th day was either clinical or related to patient age. Waiting with lengthening lasted until the swelling and pain had disappeared. In younger patients distraction was started earlier. Although the best time for starting distraction is discussible, very fast regeneration was observed in some cases of this study, even healing was detected during two weeks so that there appeared a need for a repeated bone cut. Morphologic studies by Schwartsman and Schwartsman (1992) have shown that the optimal length of this period should be seven to ten days in cases of corticotomy. Yasui and the coauthors (1993) believe that a waiting period after osteotomy is more practical than achieving immediate distraction after uncertain corticotomy. As the patients of the present study were mostly children, it proved that in cases of corticotomy an earlier start of distraction is indicated.

The lengthening index was calculated, the average for the present study being 29.5 days per centimetre. The lengthening indices were larger in congenital cases because of the affection bone reparative potency. In post-infection cases we did not observe elongation of bone regeneration time, however, this can be explained by very low patient age in epipyseal osteomyelitis cases and probably also by the fact that infection did not affect bone regeneration potency. The healing/lengthening index is used for the description of bone lengthening (Monticelli and Spinelli 1981; DeBastiani *et al* 1987), Sakurakichi and the coauthors (2002) recommend to use the indices of distraction and maturation as the components of total lengthening separately.

Calculation of the time required for the lengthening of a lower limb segment by one percent (Noonan *et al* 1998) did not seem necessesary, as comparison of the lengthening indices for groups with different etiology, techniques used, and site of the procedure gives enough valuable information about a limb lengthening procedure (Moseley 1989; Franke *et al* 1990; Guidera *et al* 1991; Guarniero *et al* 1993; Stanitski *et al* 1996; Aldegheri 1997; Aronson 1997; Yun *et al* 2000; Zarzycki *et al* 2002; Eralp *et al* 2004; Karlen *et al* 2004).

Complication rate was not high in the present study, an average of 49% in epimetaphyseal limb lengthenings, however 21% of the complications were major complications which needed further surgical treatment. A simple grading of complications into minor and major was used. Different grading systems of complications have been advocated but this simple system is more reasonable and gives enough information. The amount of lengthening at an acceptable complication rate should not exceed 25% (Maffuli and Fixsen 1996). Complication rate has varied with respect to the amount and method of limb lengthening, ranging from 22% (Guarniero *et al* 1993) and 30% (Garcia-Cimbrelo *et al* 1992) to even 1.2 major complications per patient (120%) (Guidera *et al* 1991). At the same time, it seems impossible to avoid minor complications when external fixators are used (Yeu *et al* 1994). Clinical observation of several hundreds of limb lengthening patients demonstrated prolonged muscle weakness in the involved limb after treatment. The etiology of this sign may be either neuropathic or myopathic (Young *et al* 1993) as described in electromyographic studies. This suggestion may explain joint stiffness which appears during limb lengthening.

In the present study it was found that femur lengthening damages the extensor muscles of the femur (motor units). Muscle damage is correlated to the extent of femoral lengthening but is not influenced by the etiology of limb shortening, or by the method, velocity and duration of limb lengthening. The limitations of joint function in limb lengthening disappeared during one to two years. These findings are also supported by other investigators (Barker *et al* 2001). Distraction during a limb lengthening procedure causes substantial morphological changes in the muscles and nerves (Battiston *et al* 1992; Chandler *et al* 1988; Stephens 1983; Haviko 1989). This finding was confirmed by an experimental study with rabbits, where the incidence of neuromuscular injury was high (Chuang *et al* 1995). The symptoms of such an injury to the peripheral nerves and muscles, although rather common, usually disappear when the rate of distraction is reduced, or after the procedure (Galardi *et al* 1990; Faber et al 1991; Rajacich *et al* 1992; Karger *et al* 1993). Muscle capability for lengthening is 20–30%, persistent damage to muscles may appear later (Lindsey *et al* 2002). Sofield and the coworkers (1958), and Kawamura and the coworkers (1968), and Macnicol and Catto (1982) reported gait disturbances and loss of strength as many as twenty years after a limb lengthening procedure. Similarly, it was found in the current study that on average 65% of patients suffered from limping during follow-up. Holm and the coworkers (1995) showed that limb lengthening may have a permanent effect on the neuromuscular tissue.

Posttraumatic limb shortening as the result of premature closure of the growth cartilage and severe traumas with large bone defects is a very complicated clinical problem. In earlier studies evidence was gained that posttraumatical limb shortening occupies the third place after congenital limb shortening and postinfectional limb shortening (Haviko, 1989). In minor lengthening with the correction of angular deformity, distraction epiphysiolysis with triangular or trapezium-shaped regenerates was formed. In that group of patients also closed metaphyseal distraction was applied (2 patients), which created good possibilities for the correction of deformities. At present, publications reporting closed distraction of the metaphyseal bone without corticotomy are not available. In the group of posttraumatic patients regenerative capacity was not impaired in general, as the time spent for 1 cm limb lengthening was approximately one month.

Posttraumatic limb lengthening also showed that in the case of repeated lengthening, bone reparation capability is limited. The patient who underwent femoral lengthening three times showed an increase in the lengthening index from 26.4 days per centimetre for first femoral lengthening to 46.4 for the third lengthening. According to an earlier study, the poorest functional and psychological outcome was reported in patients who underwent limb lengthening because of trauma (Tjerneström and Rehnberg 1994). Still the present study did not confirm this. Limb lengthening gained in the present study remained lower than 20% in most study groups which explains the good results.

A good result was obtained in extensive limb lengthening with a simultaneous deformity correction in Ollier's disease. Another goal was replacement of the pathological cartilagineous foci characteristic of the disease. Although similar attempts have been made by other investigators, 32 cm femoral and tibial lengthening has not been reported earlier (Urist 1989; Mitchell and Ackerman 1987; Pandey *et al* 1995). In line with the historical guidelines suggesting limits for the amount of limb lengthening, treatment results and complications have been reported for patients in whom the initial goal of lengthening exceeded 20% of initial segment length. In these cases, complication rate was 0.9 per lengthened segment, but good to excellent results were achieved in 78% of cases, which indicates that the authors had great success (Yun *et al* 2000). No complications were observed in large-amount limb lengthening in this case.

In the present study the rate of response to the questionnaire was high (70%). Yet those who did not respond (16 patients with 11 complications) did not do so not because of the poor result only but for other reasons as well. At least five of such patients are living abroad and the questionnaire did not reach them.

Despite the fact that 24 (65%) of the respondents are limping most or all of the time, 29 (78%) are satisfied with the present situation and 25 (68%) are satisfied with the cosmetic appearance. High satisfaction is demonstrated by the fact that 30 (81%) of the patients would pass limb lengthening once more if it were indicated. However, as the question “Would you do it again?” is highly emotional, most patients would agree to pass the surgery once more.

Although the pain scores for different locations were calculated, we did not observe significant statistical correlation between diagnosis, method of operation and other indicators. Only pain in the knee joint is associated with more impaired function.

Already Brockway and Fowler (1942) had good or satisfactory results in 87% of limb lengthening patients. Most patients, 88%, stated that they would undergo the treatment again if it were indicated (Ramaker *et al* 2000). According to another study, 82% of paediatric patients were satisfied with the overall result, but only 67% of them thought that the duration of the treatment was reasonable and said that they would be willing to have the procedure a second time if it were indicated (Ghoneem *et al* 1996). Patients were also more satisfied with thigh scars than with leg scars because clothing can easily cover thigh scars (Ghoneem *et al* 1996). In the present study, 25 patients were generally satisfied with the cosmetic result but 19 patients were satisfied with the appearance of scars.

There was no significant difference between the methods used in the present study – distraction epiphysiolysis showed even better functional outcome despite the fact that the procedure was performed close to the joint. Yet these patients were more dissatisfied with the result than the others. Limb lengthening from the epimetaphyseal region of the bone is still topical, especially when there is a need for a simultaneous deformity correction.

Limb lengthening requires careful planning and meticulous outpatient care and is still burdened by minor complications. Experience may lessen major complications, however, pin tract problems, swelling, and pain will plague these patients. A large group of patients can benefit from lengthening procedures, and new techniques are improving the risk-benefit balance. A thoughtful approach is needed by the surgeon, especially taking time to fully inform the patient about the time, effort, and problems involved in substantial limb.

Within this thesis experimental an clinical studies of lower limb lengthening were performed. The experimental studies demonstrated the important role of endosteal regeneration and its interaction with intramembranous regeneration. Further studies are needed for more thorough analysis of the cellular and molecular composition of the bone regenerate. The clinical studies of lower limb lengthening revealed no significant difference between the methods of operation; results differed among the etiology groups. The patients were satisfied with the long-term result. At present limb lengthening is moving towards the use of intramedullar nails. In cases when deformities are located near the joint, or involve the whole intramedullary bone canal, the methods described in this thesis will definitely be useful. Still investigations should be directed to improve operation methods and techniques. As no studies have focused on the long-term result of limb lengthening, the studied patients should undergo further follow-up.

7. CONCLUSIONS

1. Experimental bone regeneration after corticotomy in rabbit tibia was characterized by a process of early osteoclastic resorption and osteoblastic proliferation. Regeneration of the lengthened tibia was more rapid compared with the enchondral ossificaton of tibia on the control limb. Circulation of the bone regenerate was not impaired as venous hyperaemia was observed during the first week after operation.
2. Repair of a tibial defect after resection osteotomy in rats was similar to embryohistogenesis with periostal primary and endosteal secondary ossification. After bone cortex perforation, endosteal ossification followed immediately without the chondrous stage, whereas periosteal ossification occurred through the chondrous callus. In experimental animal with the isolated periost formation of the endosteal callus was inhibited and formation of the periosteal callus was mostly arrested.
3. Limb lenthening damaged the extensor muscles of the knee (motor units), the amount of damage varied according to the extent of the lengthening of the femur. Muscle damage was not related to the aetiology of limb shortening and to the method, rate and duration of limb lengthening. The vastus medialis muscle was more affected than the rectus femoris and vastus lateralis muscles.
4. In limb lengthening after posttraumatic premature closure of the growth cartilage and after severe traumas with large bone defects, four different methods of limb lengthening were used depending on the grade of damage. Ossification of the bone regenerate was not disturbed and the rate of complications was low. Multiple limb lengthening may act on bone regeneration capacity.
5. In an evaluated case of Ollier's disease extensive limb segment lengthening (75% of the initial length of the femur) was effective through the osteochondral foci of the disease. During this procedure the osteochondral foci with embryonic cartilage cells were replaced with the bone tissue. Altogether 32 cm limb lengthening was gained, while no complications were detected. The patient was satisfied with the treatment result.
6. Distraction limb lengthening was performed in 53 patients from the epimetaphyseal region, while mostly metaphyseal corticotomy (57%) or osteotomy (26%) was performed using a special osteotome. A mean 26.2% (range 8.5–81) of lower limb lengthening was gained. The mean lengthening index was 29.5 (range 12.9–59.4) days per centimetre.
7. Long-term follow-up (mean 19.4 years) revealed that 78% of the patients were satisfied with their present situation and 68% were satisfied with the cosmetic appearance, which depended on complications as well as patient's gender. Although 81% of the patients agreed to pass limb lengthening once more if were needed; this decision was not correlated with the objectively estimated functional indicators.

8. REFERENCES

Aarnes GT, Steen H, Kristiansen LP, Ludvigsen P, Reikerås O. Tissue response during monofocal and bifocal leg lengthening in patients. J Orthop Res 2002; 20: 137–41
Abbott LC The operative lengthening of the tibia and fibula. J Bone Joint Surg 1927; 9: 128–52
Aldegheri R, Trivella G, Renzi-Brivio L, Tessari G, Agostini S, Lavini F. Lengthening of the lower limbs in achondroplastic patients. A comparative study of four techniques. J Bone Joint Surg 1988; 70B (1): 69–73
Aldegheri R, Renzi-Brivio L, Agostini S. The callotasis method of limb lengthening. Clin Orthop 1989; 241: 137–45
Aldegheri R. Callotasis J Pediatr Orthop 1993; B2(1): 11–5
Aldegheri R. Femoral callotasis J Pediatr Orthop 1997; B6(1): 42–7
Aldegheri R. Distraction osteogenesis for lengthening of the tibia in patients who have limb length discrepancy or short stature. J Bone Joint Surg 1999; 81A: 624–34
Alho A, Bang G, Karaharju E, Armond I. Filling of a bone defect during experimental osteotaxis distraction. Acta Orthop Scand 1982; 53: 29–34
Aronson J. Experimental and clinical experience with distraction osteogenesis. Cleft Palate Craniofac J 1994; 31: 473–82
Aronson J, Shen X. Experimental healing of distraction osteogenesis comparing metaphyseal with dipyseal sites Clin Orthop 1994; 301: 25–30
Aronson J. Limb lengthening, skeletal reconstruction, and bone transport with the Ilizarov method. J Bone Joint Surg 1997; 79A; 1243–58
Aronson J, Gao GG, Shen XC, McLaren SG, Skinner RA, Badger TM and Lumpkin CK, Jr. The effect of aging on distraction osteogenesis in the rat. J Orthop Res 2001; 19: 421–7
Barker KL, Simpson AHRW, Lamb SE. Loss of knee range of motion in leg lengthening. J Orthopaedic & Sports Physical Therapy 2001; 31(5): 238–44
Battiston B, Buffoli P, Vigasio A, Brunelli G, Antonini L. The effects of lengthening on nerves. Italian J Orthop and Traumatol 1992; 18: 79–86
Baumgart R, Kettler M, Zeiler C, Betz A, Schweiberer L. Möglichkeiten der knochendurchtrennung. Osteotomie- und kortikotomietechniken. Unfallchir. 1997; 100: 797–804
Bertram C, Nieländer K-H, König DP. Pioniere der Extremitätenverlängerung. Chirurg 1999; 70: 1374–8
Bessler W, Grauer W, Allemann J. Case report 726. Skeletal Radiol 1992; 21:201–4
Bigland-Ritchie B, Cafarelli E,Vollestad NK. Fatigue of submaximal static contractions. Acta Physiol Scand 1986; Suppl 556: 137–48.
Birch JG, Samchukov ML. Use of the Ilizarov method to correct lower limb deformities in children and adolescents. Journal of the American Academy of Orthopaedic Surgeons 2004; 12 (3): 144–54
Bjerkreim I. Limb lengthening by physeal distraction. Acta Orthop Scand 1989; 60(2): 140–2
Bliskunov AI. Intramedullary distraction of the femur. Orthop Travmatol Protez 1983; 10: 59–62 (rus)
Blane CE, Herzenberg JE, DiPietro MA. Radiographic imaging for Ilizarov limb lengthening in children. Pediatr. Radiol 1991; 21(2): 117–20

Bonnard C, Favard L, Sollogub I, Glorion B. Limb lengthening in children using the Ilizarov method. Clin Orthop. 1993; 293: 83–8

Bowen JR, Levy EJ, Donohue M. Comparison of knee motion and callus formation in femoral lengthening with the Wagner or monolateral-ring device. J Pediatr Orthop 1993; 13(4): 467–72

Brockway A, Fowler SB: Experiences with 105 leg-lengthening operations. Surg Gynecol Obstet 1942; 72:252–6

Brug E, Klein W, Baranowski D, Winckler S. August Bier-ein Pionier der Kallusdistraktion. In: Wolter D, Zimmer W, eds. Die Plattenosteosynthese und ihre Konkurrenzverfahren. Berlin: Springer. 1991: 47–52

Carnesale PG. Multiple Enchondromatosis (Ollier Disease). In: Canale ST, ed. Campbell's Operative Orthopaedics 10^{th} ed. St.Louis: Mosby. 2003 Vol 1: 809

Catagni MA, Lovisetti L, Guerreschi F, Combi A, Ottaviani G. Cosmetic bilateral leg lengthening. Experience of 54 cases. J Bone Joint Surg 2005; 87–B(10): 1402–5

Cech O, Trc T. Prof. Ilizarov and his contribution to the challenge of limb lengthening. Injury 1993; 24 (Suppl 2): 2–7

Chambers TJ. The cellular basis of bone resorption. Clin Orthop 1980; 157: 283–93

Chandler D, King JD, Bernstein SM, Marrero G, Koh J, Hambrecht H. Results of 21 Wagner limb lengthenings in 20 patients. Clin Orthop 1988; 230: 214–22.

Choi IH, Chung CY, Cho T-J, Yoo WJ. Angiogenesis and mineralization during distraction osteogenesis. J Korean Med Sci 2002; 17: 435–47

Chuang TY, Chan RC, Chin LS, Hsu TC. Neuromuscular injury during limb lengthening: a longitudinal follow-up by rabbit tibial model. Archives of Physical Medicine & Rehabilitation. 1995; 76(5): 467–70

Codivilla A. On the means of lengthening, in the lower limbs, the muscles and tissues which are shortened through deformity. Am J Orthop Surg 1905; 2: 353–69

Coleman SS, Scott SM. The present attitude toward the biology and technology of limb lengthening. Clin Orthop 1991; 264: 76–83

Dahl MT, Gulli B, Berg T. Complications of limb lengthening. A learning curve. Clin. Orthop. 1994; 301:10–8

De Bastiani G, Aldegheri R, Renzi Brivio L, Trivella G. Limb lengthening by distraction of the epiphyseal plate. A comparison of two techniques in the rabbit. J Bone Joint Surg 1986; 68Br: 545–9

De Bastiani G, Aldegheri R, Renzi Brivio L, Trivella G. Chondrodiatasis – controlled symmetrical distraction of the epiphyseal plate. Limb lengthening in children. J Bone Joint Surg 1986; 68Br(4): 550–6

De Bastiani G, Aldegheri R, Renzi-Brivio L, Trivella G. Limb lengthening by callus distraction (callotasis). J Pediatr Orthop 1987; 7: 129–34

DePablos J, Villas C, Canadell J. Bone lengthening by physeal ditraction. An experimental study. Int Orthop 1986; 10: 163–70

Desai M. P. Growth Disorders. MJAFI (Medical Journal Armed Forces of India) 2003; 59 : 278–82

Dickson FD, Diveley RL. A new apparatus for the lengthening of legs. J Bone Joint Surg 1932; 14:194–6

Donnan LT, Saleh M, Rigby AS, McAndrew A. Radiographic assessment of bone formation in tibia during distraction osteogenesis. J Pediatr Orthop 2002; 22(5): 645–51

Dusmuratov MD, Dusmuratov MM. Effect of some factors on the distraction epiphyseolysis. Ortoped Travmatol Protez 1984; 9: 30–3 (rus)

Dutoit M, Rigault P, Padovani J, Finidori G, Touzet HP, Durand Y. The outcome for children treated by limb lengthening for congenital hypoplasia of the lower limbs. Rev Chir Orthop 1990; 76: 1–7

Einhorn TA. Enhancement of fracture healing. J Bone Joint Surg 1995; 77A: 940–56

Eitel F, Seiler H, Schveiberer L. Vergleichende morphologische untersuchungen zur übertragbarkeit für experimentelle ergebnisse auf den regenerationsprozess des menschlichen rörenknochens. II untersuchungsergebnisse. Unfallheilkunde 1981; 84: 255–64

Eldridge JC, Bell DF. Problems with substantial limb lengthening. Orthop Clin North America 1991; 22: 625–31

Eralp L, Kocaoglu M, Ozkan K, Turker M. A comparison of two osteotomy techniques for tibial lengthening. Arch Orthop Trauma Surg 2004; 124(5): 298–300

Faber FW, Keessen W, van Roermund PM. Complications of leg lengthening. 46 procedures in 28 patients. Acta Orthop Scand 1991; 62: 327–32

Fink B, Fox F, Singer J, Skripitz R, Feldkamp J. Monitoring of bone formation during distraction osteogenesis via osteocalcin: a time sequence study in dogs. J Orthop Sci 2002; 7: 557–61

Fischenko PJ, Sadofieva VI, Karimova LF, Pilipenko NP. Roentgenologic imaging of the regenerate formation in distraction epiphysiolysis. Ortoped Travmatol Protez 1976; 11: 29–33

Fischgrund J, Paley D, Suter C. Variables affecting time to bone healing during limb lengthening. Clin Orthop 1994; 301: 31–7

Fjeld TO, Steen H. Growth-retardation after experimental limb lengthening by epiphyseal distraction. J Pediatr Orthop 1990; 10(4): 463–6

Franke J, Hein G, Simon M, Hauch S. Comparison of distraction epiphyseolysis and partial metaphyseal corticotomy in leg lengthening. Intl Orthop 1990; 14: 405–13

Galardi G, Comi G, Lozza L, Marchettini P, Novarina M, Facchini R, Paronzini A. Peripheral nerve damage during limb lengthening. Neurophysiology in five cases of bilateral tibial lengthening. J Bone Joint Surg 1990; 72B(1): 121–4

Garcia-Cimbrelo E, Olsen B, Ruiz-Yague M, Fernandez-Baillo N, Munuera-Martinez L. Ilizarov technique. Results and difficulties. Clin Orthop 1992; 283: 116–23

Garcia-Cimbrelo E, Mano A, Curto DL, Garcia-Rey E, Cordero J, Marti-Ciruelos R. The intramedullary elongation nail for femoral lengthening. J Bone Joint Surg 2002; 84B(7): 971–7

Ghoneem H, Wright J, Cole WG, Rang M. The Ilizarov method for correction of complex deformities. Psychological and functional outcomes. 1996; 78A(10): 1480–5

Glorion C, Pouliquen JC, Langlais J, Ceolin JL, Kassis B. Femoral lengthening by callotasis. A study of a series of 79 cases in children and adolescents. Rev Chir Orthop Reparatrice Appar Mot 1995; 81:147–56

Goransson H, Pätiälä H, Linden M, Vuola J, Rokkanen P. Histology and histomorphometry of bone regeneration after experimental injuries. Ann Chir Gyn 1992; 81: 58–65

Gordh M, Alberius P, Linberg L, Johnell O. Bone graft incorporation after cortical perforations of the host bed. Otolaryngol Head Neck Surg 1997; 117: 664–70

Grundnes O, Reikeras O. Effect of physical activity on muscle and blood flow after fracture. Exercise and tenotomy studied in rats. Acta Orthop Scand 1991a; 62: 67–9
Grundnes O, Reikeras O. Mechanical effects of function on bone healing. Nonweight bearing and exercise in osteotomized rats. Acta Orthop Scand 1991b; 62: 163–5
Guarniero R, Montenegro NB, Guarnieri MV, Rossi JD. Comparative-study of Ilizarov, Wagner, and Anderson methods for limb lengthening (100 lengthenings in 98 patients). J Pediatr Orthop 1993; B2(1): 28–34
Guerino MR, Luciano E, Goncalves M, Leivas TP. Effects of chronic physical activity and ultrasound treatment on bone consolidation. Physiol Chem Phys Med NMR 1999; 31: 131–8
Guichet JM, Braillon P, Bodenreider O, Lascombes P. Periosteum and bone marrow in bone lengthening: a DEXA quantitative evaluation in rabbits. Acta Orthop Scand 1998; 69(5): 527–31
Guidera KJ, Hess WF, Highhouse KP, Ogden JA. Extremity lengthening: results and complications with the Ortofix system. J Pediatr Orthop 1991; 11(1): 90–4
Guidera KJ. Helal AA. Zuern KA. Management of pediatric limb length inequality. Advances in Pediatrics. 1995; 42: 501–43
Gyulnazarova SV, Shtin V.P. Reparative bone tissue regeneration in treatment of pseudoarthroses with simultaneous elongation in the pathologic focus region. Ortoped Travmatol Protez 1983; 4: 10–5 (rus)
Hagg GM. Comparison of different estimators of electromyographic spectral shifts during work when applied on short test contractions. Med Biol Eng Comput 1991; 29: 511–6.
Hamanishi C, Yasuwaki Y, Kikuchi H, Tanaka S, Tamura K. Classification of the callus in limb lengthening – radiographic study of 35 limbs. Acta Orthop Scand 1992; 63(4): 430–3
Hamdy RC, Silvestri A, Rivard CH, Ehrlich M. Histologic evaluation of regenerate bone in cases of limb lengthening by the Ilizarov technique. An experimental study in dogs. Ann Chir 1997; 51(8): 875–83
Hankemeier S, Bastian L, Gosling T, Krettek C. Prinzipen der kallusdistraktion. Unfallchirurg 2004; 107: 945–59
Hankemeier S, Gosling T, Pape HC, Wiebking U, Krettek C. Limb lengthening with the intramedullary skeletal kinetic distractor (ISKD). Oper Orthop Traumatol 2005; 17:79–101.
Hantes ME, Malizos KN, Xenakis TA, Beris AE, Mavrodontidis AN, Soucacos PN. Complications in limb-lengthening procedures: a review of 49 cases. Am J Orthop 2001; 30(6): 479–83
Haviko T. Closed distraction epiphyseolysis. Thesis. Moscow 1977(rus)
Haviko T. Surgical correction of limb shortening and deformations. Doctoral thesis. Riga Medical Institute, Riga, Latvia, 1989 (rus)
Heikkinen E, Vihersaari T, Penttinen R. Effect of previous exercise on fracture healing: a biochemical sudy with mice. Acta Orthop Scand 1974; 45: 481–9
Herzenberg JE, Scheufele LL, Paley D, Bechtel R, Tepper S. Knee range of motion in isolated femoral lengthening. Clin Orthop 1994; 301: 49–54
Holm I, Steen H, Ludvigsen P, Bjerkreim I. Unchanged muscle function after bilateral femoral lengthening. A prospective study of 9 patients with a 2-year follow-up. Acta Orthop Scand 1995; 66(3): 258–60

Hrutkay JM, Eilert RE. Operative lengthening of the lower extremity and associated psychological aspects: the Children's Hospital experience. J Pediatr Orthop 1990; 10(3): 373–7
Hughes TH, Maffulli N, Green V, Fixsen JA. Imaging in bone lengthening. A review. Clin Orthop 1994; 308: 50–3
Ilizarov GA, Soibelman LM. Some clinical and experimental data concerning bloodless lengthening of lower extremities. Eksper Khir Anesteziol 1969; 4: 27–32 (rus)
Ilizarov GA, Ledyaev VI, Shitin VP. The course of reparative regeneration of the compact bone in distraction osteosynthesis under different conditions of fixation of bone fragments (an experimental study). Eksper Khir Anesteziol 1969; 6: 3–12 (rus)
Ilizarov GA, Deviatov AA. Operative elongation of the leg. Ortoped Travmatol Protez 1971; 8: 20–5 (rus)
Ilizarov GA, Trohova VG. Operative elongation of the hip. Ortop Travmatol Protez 1973; 11: 51–5 (rus)
Ilizarov GA, Khelimsky AM, Deviatov AA, Kataev IA, Ivanov GG. Reparative regeneration of the bone tissue during replacement of defects of long bones by lengthening one of the fragments. Eksper Khir Anesteziol 1975; 2: 37–42 (rus)
Ilizarov GA, Helimsky AM, Barabash AP. Dependence of course of reparative osteogenesis in experimental elongating arthrodesis of the knee joint on the terms of start of the distraction. Ortoped Travmatol Protez 1977; 2: 23–8 (rus)
Ilizarov GA, Schreiner AA. A new method of closed flexion osteoclasia (experimental study). Ortoped Travmatol Protez. 1979; 40: 9–14 (rus)
Ilizarov GA, Berko VG. Morphologic characteristic of the regenerate, formed in elongation of the hip in experiment. Ortoped Travmatol Protez 1980; 7: 54–9 (rus)
Ilizarov GA, Barabash AP, Imerlishvili IA, Larionov AA, Kochetkov Y.S. Morphologic characteristic of bone tissue formation and reconstruction in the substitution of large bone defect. Ortoped Travmatol Protez. 1984; 1: 16–20 (rus)
Jani L. Die distraktionsepiphyseolyse. Tierexperimentelle studie zum problem der beinverlängerung Teil I. Z Orthop 1975; 113: 189–98.
Johnson MA, Polgar J, Weightman D, Appleton D. Data on the distribution of fibre types in thirty-six human muscles. An autopsy study. J Neurol Sci 1973; 18: 111–29
Karger C, Guille JT, Bowen JR. Lengthening of congenital lower limb deficiencies. Clin Orthop 1993; 291: 236–45
Karlen LKP, Yinusa W, Yan LS, Wang KW, Hoi LY, John LCY. Analysis of scar formation after lower limb lengthening – Influence on cosmesis and patient satisfaction. J Pediatr Orthop 2004; 24(6): 706–10
Kawamura B, Hosono S, Takahashi T, Yano T, Kobayashi Y, Shibata N, Shinoda Y. Limb lengthening by means of subcutaneous osteotomy. Experimental and clinical studies. J Bone Joint Surg 1968; 50A: 851–78.
Kojimoto H, Yasui N, Goto T, Matsuda S, Shimomura Y. Bone lengthening in rabbits by callus distraction. J Bone Joint Surg 1988; 70B: 543–9
Kolbeck S, Bail H, Weiler A, Windhagen H, Haas N, Raschke M. Digital radiography: a predictor of regenerate bone stiffness in distraction osteogenesis. Clin Orthop 1999; 366: 221–8
Korzinek K, Tepic S, Perren SM. Limb lengthening and three-dimensional deformity corrections. Arch Orthop Trauma Surg 1990; 109: 334–40

Kristiansen PL, Steen H. Reduced lengthening index by use of bifocal osteotomy in the tibia: comparison of monofocal and bifocal procedures with the Ilizarov external fixator. Acta Orthop Scand 2002; 73: 93–7

Krompecher S. Die Beeinflussbarkeit der Gewebsdifferenzierung der granulierenden Knochenoberflächen, insbesondere die der Callusbildung. Langenbecks Arch Klin Chir 1956; 281: 472–512

Landry PS, Marino AA, Sadasivan KK, Albright JA. Bone injury response. An animal model for testing theories of regulation. Clin Orthop 1996; 332: 260–73

Langlois V, Laville JM. Physeal distraction for limb length discrepancy and angular deformity. Rev Chir Orthop

Reparatrice Appar Mot 2005; 91:199–207

Lavrishtsheva GI, Dubrov E.Y. Value of compression and diastasis between fragments in union of bones by first intention. Ortoped Travmatol Protez 1968; 8: 9–12 (rus)

Ledyaev VI. Regeneration of the bone tissue of the diaphysis under different conditions of osteosynthetic traction under experimental conditions. Eksper Khir Anesteziol 1975; 2: 42–9 (rus)

Lee SH, Szoke G, Simpson H. Response of the physis to leg lengthening. J Pediatr Orthop 2001; 10B(4): 339–43

Lenzner A. Treatment of pelvic fractures. Thesis. Riga 1986 (rus)

Li G, Simpson AHRW, Triffitt JT. The role of chondrocytes in intramembranous and endochondral ossification during distraction osteogenesis in the rabbit. Calcif Tissue Int 1999; 64: 310–7

Lindsey CA, Makarov MR, Shoemaker S, Birch JG, Buschang PH.; Cherkashin AM, Welch RD, Samchukov ML. The effect of the amount of limb lengthening on skeletal muscle. Clin Orthop 2002; 402: 278–87

Lindstrom L, Magnusson R, Petersen I. Muscular fatigue and action potential conduction velocity changes studied with frequency analysis of EMG signals. Electromyography 1970; 10: 341–56

Little C. Ollier disease: an interdisciplinary approach. Orthopaedic Nursing 1994; 13: 50–5

Macnicol MF, Catto AM. Twenty-year review of tibial lengthening for poliomyelitis. J Bone Joint Surg 1982; 64B(5): 607–11

Maffuli N, Fixsen JA. Distraction osteogenesis in congenital limb length discrepancy: a review. Journal of the Royal College of Surgeons of Edinburgh. 1996; 41(4): 258–64

Merle d'Aubigne R, Dubousset J. Surgical correction of large length discrepancies in the lower extremities of children and adults. An analysis of twenty consecutive cases. J Bone Joint Surg 1971; 53A(3): 411–30

Mitchell ML, Ackerman LV. Case report 405. Skeletal Radiol 1987; 16: 61–66

Monticelli G, Spinelli R. Distraction epiphysiolysis as a method of limb lengthening. Clin Orthop 1981; 154: 274–85

Monticelli G, Spinelli R. Leg lengthening by closed metaphyseal corticotomy. Ital J Orthop Traumatol 1983; 9(2): 139–50

Moore BH: A critical appraisal of the leg lengthening operation. Am J Surg 1941; 52:415–23

Moseley CF. Leg lengthening. A review of 30 years. Clin Orthop 1989; 247: 38–43

Müller ME, Nazarian S, Koch P. Classification AO des fractures. Berlin-Heidelberg-New-York: Springer –Verlag. 1987

Noonan K, Leyes M, Forriol F, Canadell J. Distraction osteogenesis of the lower extremity with use of monolateral external fixator. A study of two hundred and sixty one femora and tibiae. J Bone Joint Surg 1998; 80A: 793–806

O'Beirne J, Maher M, O'Flanagan S, McGuinness A. Early experience with leg lengthening by callotasis and chondrodiastasis. Ir J Med Sci 1993; 162: 301–5

Paley D. Current techniques of limb lengthening. J Pediatr Orthop 1988; 8: 73–92

Paley D. Problems, obstacles, and complications of limb lengthening by the Ilizarov technique. Clin Orthop 1990; 250: 81–104

Pandey R, White SH, Kenwright J. Callus distraction in Ollier's disease. Acta Orthop Scand 1995; 66: 479–80

Paterson D. Leg-lengthening procedures: A historical review. Clin Orthop 1990; 250: 27–33

Peltonen J, Alitalo I, Karaharju E, Heliö H. Distraction of the growth plate. Experiments in pigs and sheep. Acta Orthop Scand 1984a; 55: 359–62

Peltonen J, Karaharju E, Alitalo I. Experimental epiphysial distraction producing and corrcting angular deformities. J Bone Joint Surg 1984b; 66B: 598–602

Pflüger G, Rahn BA, Fischerleitner F, Thoma H, Wolner Ch. Knöcherne auffüllung des distraktionsspaltes bei beinverlängerung. Arch Orthop Unfall-Chir 1976; 86: 45–60

Poljanski V. External fixation of open tibial shaft fractures. Thesis. Moscow 1986 (rus)

Pouliquen JC, Chaboche P, Pennecot GF, Bergue A. Etude experimentale du retentissiment sur le cartilage de croissance et les parties molles de l'allongement progressif du femur chez le lapin en periode de croissance. Chir Pediat 1980; 21: 363–7

Putti V. The operative lengthening of the femur. JAMA 1921; 77: 934–5

Rajacich N, Bell DF, Armstrong PF. Pediatric applications of the Ilizarov method. Clin Orthop 1992; 280: 72–80

Ramaker RR, Lagro SWJ, van Roermund PM, Sinnema G. The psychological and social functioning of 14 children and 12 adolescents after Ilizarov leg lengthening. Acta Orthop Scand 2000; 71(1): 55–9

Reichel H, Lebek S, Alter C, Hein W. Biomechanical and densitometric bone properties after callus distraction in sheep. Clin Orthop 1998; 357: 237–46

Remedios A. Bone and bone healing. Vet Clin North Am Small Anim Pract 1999; 29: 1029–44

Roesler H., Rompe G. Beinlängendifferenz und verkürzungsausgleich. Z Orthop 110 (1972) 623–628

Sabharwal S, Paley D, Bhave A, Herzenberg JE. Growth patterns after lengthening of congenitally short lower limbs in young children. J Pediatr Orthop 2000; 20(2): 137–45

Sakurakichi K, Tsuchiya H, Uehara K, Kabata T, Tomita K. The relationship between distraction length and treatment indices during distraction osteogenesis. J Orthop Sci 2002; 7: 298–303

Saleh M, Goonatillake HD Management of congenital leg length inequality: value of early axis correction. J Pediatr Orthop B 1995; 4: 150–58

Saleh M, Hamer AJ. Bifocal limb lengthening – a preliminary report. J Pediatr Orthop 1993; B2(1): 42–8

Schwartsman V, Schwartsman R. Corticotomy. Clin Orthop. 1992; 280: 37–47

Shtin VP, Nikitenko TT. Basing the term of beginning of distraction operative lengthening of the leg in experiment. Ortop Travmatol Protez 1974; 5: 48–51 (rus)

Shtin VP. Gyulnazarova SV, Nikitenko ET, Chirkova AM. Time of commencement of distraction in the diaphyseal elongation of the long tubular bones (Experimenal study). Ortoped Travmatol Protez 1979; 1: 14–8 (rus)

Shugarov NA, Arapov NA. Effect of pins on the blood-vascular system of bone. Ortoped Travmatol Protez 1975; 10: 66–8 (rus)

Simpson H. Barker K. Effect on knee flexion of a modification to the surgical technique of pin placement during femoral lengthening. J Pediatr Orthop B 2002; 11(4): 307–12

Sofield HA, Blair SJ, Millar EA. Leg-lengthening. A personal follow-up of forty patients some twenty years after the operation. J Bone Joint Surg 1958; 40A: 311–22

Song HR, Puri A, Lee JH, Park HB, Ra DK, Kim GS, Yeon SC. Spontaneous bone regeneration in surgically induced bone defects in young rabbits. J Pediatr Orthop B, 2002; 11(4): 343–9

Solomonow M, Baten C, Smit J, Baratta R, Hermens H, D'Ambrosia R, Shoji H. Electromyogram power spectra frequencies associated with motor unit recruitment strategies. J Appl Physiol 1990; 68: 1177–85

Stanitski DF, Bullard M, Armstrong P, Stanitski CL. Results of femoral lengthening using the Ilizarov technique. J Pediatr Orthop 1995; 15: 224–31

Stanitski DF, Rossman K, Torosian M. The effect of femoral lengthening on knee articular cartilage: The role of apparatus extension across the joint. J Pediatr Orthop 1996; 16(2): 151–54

Startseva IA, Gorbunova ZI. Effect on the distraction epiphysiolysis on the growth of the elongated bone (experimental study). Ortop Travmatol Protez 1982; 6: 36–41 (rus)

Steen H. Fjeld TO. Lengthening osteotomy in the metaphysis and diaphysis. An experimental study in the ovine tibia. Clin Orthop 1989; 247: 297–305

Steen H. Fjeld TO. Miller JA. Ludvigsen P. Biomechanical factors in the metaphyseal- and diaphyseal-lengthening osteotomy. An experimental and theoretic analysis in the ovine tibia. Clin Orthop 1990; 259: 282–94

Stephens DC. Femoral and tibial lengthening. J Pediat Orthop 1983; 3: 424–30

Stulen FB, DeLuca CJ. Frequency parameters of the myoelectric signal as a measure of muscle conduction velocity. IEEE Trans Biomed Eng 1981; 28: 515–23

Sveshnikov AA, Marhashov AM, Gracheva VI. Role of circulation in the reparative osteogenesis. Ortoped Travmatol Protez 1985; 5: 23–7 (rus)

Taillard W. External fixaton – past, present, future. Introduction to the Tenth International Conference on Hoffmann external fixation. Orthopedics 1984; 7(3): 398–401

Tay BK, Le AX, Gould SE, Helms JA. Histochemical and molecular analyses of distraction osteogenesis in a mouse model. J Orthop Res 1998; 16: 636–42

Tachdjian MO Pediatric Orthopedics. 2nd ed. Philadelphia: Saunders W.B. 1990 Vol.2:1195–98

Tjernström B, Rehnberg L. Back pain and arthralgia before and after leg lengthening. Acta Orthop Scand 1994; 65(3): 328–32

Tuukanen J, Peng Z, Väänänen HK. The effect of training on the recovery from immobilization-induced bone loss of rats. Acta Physiol Scand 1992; 145: 407–11

Urist MA. 37-year follow-up of multiple stage femur and tibia lengthening in dyschondroplasia (enchondromatosis) with a net gain of 23.3 centimeters. Clin Orthop 1989; 242: 137–57

Vizkelety TL, Marschalko P. Limb lengthening operations. Acta Chir Hung 1993; 33: 55–77

Wagner H. Operative Beinverlängerung. Der Chirurg 1971; 6: 260–6
Walker CW, Aronson J, Kaplan PA, Molpus WM, Seibert JJ. Radiologic evaluation of limb- lengthening procedures. American Journal of Roentgenology (AJR). 1999; 156: 353–8
Wasserstein IS. Distraction-compression method for limb lengthening by use cylindrical cortical allograft. Riga, 1971 (rus)
Wasserstein I. Twenty-five years experience with lengthening of shortened lower extremities using cylindrical allografts. Clin Orthop 1990; 250: 150–3
Watts J. China's cosmetic surgery craze. Leg-lengthening operations to fight height predjudice can leave patients crippled. Lancet 2004; 363(9413): 958
Welch RD, Birch JG, Makarov MR, Samchukov ML. Histomorphometry of distraction osteogenesis in a caprine tibial lengthening model. J Bone Min Res 1998; 13(1): 1–9
White SH, Kenwright J. The importance of delay in distraction of osteotomies. Orthop Clin North Am 1991; 22(4): 569–79
Wiedemann M. Callus distraction: a new method?: a historical review of limb lengthening. Clin Orthop 1996; 327: 291–304
Williams P, Simpson H, Kyberd P, Kenwright J, Goldspink G. Effect of rate of distraction on loss of range of joint movement, muscle stiffness, and intramuscular connective tissue content during surgical limb-lengthening: A study in the rabbit. Anatomical Record 1999; 255(1): 78–83
Wilson JW. Vascular supply to normal bone and healing fractures. Semin Vet Surg (Small anim) 1991; 6: 26–38
Yasui N, Kojimoto H, Sasaki K, Kitada A, Shimizu H, Shimomura Y. Factors affecting callus distraction in limb lengthening. Clin Orthop 1993; 293: 55–60
Yasui N, Sato M, Ochi T, Kimura T, Kawahata H, Kitamura Y, Nomura S. Three modes of ossification during distraction osteogenesis in the rat. J Bone Joint Surg 1997; 79–B: 824–30
Yasui N, Nakase T, Kawabata H, Shibata T, Helland P, Ochi T. A teachinque of percutaneous multidrilling osteotomy for limb lengthening and deformity correction. J. Orthop Sci 2000; 5: 104–7
Yeu K, Huang SC, Liu TK. Treatment of malunion of the lower extremities by the Ilizarov technique. Formos Med Assoc 1994; 93: 403–10
Young NL, Davis RJ, Bell DF, Redmond DM. Electromyographic and nerve conduction changes after tibial lengthening by the Ilizarov method. J Pediatr Orthop 1993; 13: 473–7
Young JW, Kovelman H, Resnik CS, Paley D. Radiologic assessment of bones after Ilizarov procedures. Radiology 1990; 177: 89–93
Yun AG. Severino R. Reinker K. Attempted limb lengthenings beyond twenty percent of the initial bone length: results and complications. J Pediatr Orthop 2000; 20(2): 151–9
Zarzycki D, Tesiorowski M, Zarzycka M, Kacki W, Jasiewicz B. Long-term results of lower limb lengthening by physeal distraction. J Pediatr Orthop 2002; 22(3): 367–70
Zavyalov PV, Plaksin IT. Distraction epiphysiolysis for lengthening of the lower limbs in children. Khirurgija 1968; 7: 121–3 (rus)
Zavyalov PV, Plaksin IT. Elongation of crural bones in children using a method of distraction epiphysiolysis. Vestn Khir Grekov 1969; 103(7): 67–70 (rus)

SUMMARY IN ESTONIAN

Alajäseme pikendamine: luuregeneratsiooni eksperimentaalsed uuringud ja kliinilised kaugtulemused

Organismi kasvamine on olnud läbi aegade müsteerium ning kaua on otsitud mooduseid ja võimalusi, kuidas kasvu ja kasvamist mõjutada. Jäseme, eriti alajäseme ühepoolne lühenemine (harvem ka pikenemine) kutsub esile kaskaadi järgnevaid kompensatoorseid deformatsioone. Kuni 2 cm jala lühenemist, peamiselt küll kaasasündinud või varases lapseeas omandatud haiguse tõttu, kompenseerib täiskasvanu vaagnakalde ja lülisamba kõverdamisega. Jala lühenemist 5 cm hakkab patsient korrigeerima püstpöiaga. Veelgi suuremal lühenemisel, 7–8 cm, lisandub terve jala kõverdamine põlvest, mis omakorda võib tekitada põlveliigese painutuskontraktuuri. Tänapäeval, operatsioonitehnika ja -meetodite arenedes, on tekkinud isegi alajäsemete kosmeetilise pikendamise nõudlus ning see hakkab mõningal määral muutuma probleemiks Kaug-Ida maades.

Jäsemepikendamise meetodite ajalugu on olnud unustatud ja taasavastatud ravimeetodite lugu. Jäsemepikenduse alustajaks peetakse Alessandro Codivillat, kes publitseeris oma vaatlused 1905. aastal: tema kasutas pikenduseks skelettekstensiooni. Uuele tasandile jõudis jäsemepikendus, kui Vittorio Putti võttis kasutusele 1921 aastal välisfiksaatori, mis on viinud lõpuks distraktsioonosteogeneesi teooriate selgituse ja põhjenduseni. Uut tüüpi välisfiksaatori, varraste ja rõngastega (sisuliselt Ilizarovi aparaadi eellane) konstrueeris Wittmoser 1941. aastal. Gavril Ilizarovi esimesed tööd on ilmunud 1951 aastal, kuid jäsemepikendamise, distraktsioonosteogeneesi ja kortikotoomia probleeme on ta oma töödes käsitlenud alles alates 1960. aastate lõpust. Varasemad operatsioonimeetodid, kus kasutati periosti luult laialdast lahtiprepareerimist, ei andnud häid tulemusi ja ei ole enam kasutuses. Ilizarovi aparaat ja meetodid on läänemaailmas laialdasemalt kasutuses 1980. aastate lõpust. Tartu Ülikooli Kliinikumis oli Ilizarovi aparaadi ja meetodite kasutamine laialdane juba 1970. aastate keskpaigast.

Vastuoluliseks on jäänud arutlused endosti ja periosti suhte üle, s.t. milline on kummagi osa luu regeneratsioonis ja kuidas nad koos toimivad. Eksperimentaalses ortopeedias on tõestatud küülikute ja rottide sobivust luuregeneratsiooni uuringutesse ja tulemuste sobivust, et teha järeldusi humaanmeditsiini jaoks. Ilizarov koos kaasautoritega on tõestanud erinevatel jäseme segmentidel ja luudel luusisese verevarustuse ja endostaalse luuregeneratsiooni tähtsust. Samas on ka vastakaid uurimusi, kus on demonstreeritud, et distraktsioonosteogeneesis on oluline ainult periost.

Jäseme pikendamine on pikaajaline protsess ja tulemused on sõltuvuses patsiendi vanusest, jäseme lühenemust tekitanud patoloogiast, jäseme lühenemuse määrast ja segmendist (reis või säär), aga ka operatsioonimeetodist.

Luuregeneratsiooni hindamiseks on võetud kasutusele pikendus- ehk paranemisindeks, mis näitab, mitu päeva keskmiselt kulub ühe sentimeetri jäseme pikendamiseks. Üldjuhul jääb indeks 30–60 vahele. Lastel on ta tavaliselt väiksem kui täiskasvanutel. Kaasasündinud patoloogia korral pikendusisindeks suureneb, samuti on indeks suurem sääre pikendamisel. Jäseme pikendamise jälgimine on olnud kliiniline ja röntgenoloogiline, kuid on kasutatud ka ultraheli, kompuutertomograafilist ja DXA monitoorimist. Siiski on leitud, et fiksaatori eemaldamine jääb ikkagi arsti otsustada ning määravaks saavad kliinilised testid ja kogemus. Tüsistuste hulk on jäseme pikendamisel suur ja mõnes analüüsis on see isegi olnud üle 100% s.t. ühel patsiendil on olnud keskmiselt rohkem kui üks tüsistus. Tüsistuste hulk väheneb kogemuste kasvades ja on peaaegu postuleeritud, et jäsemepikenduse operatsioonide läbiviimine peaks jääma ainult kogenud ortopeedile. Paljud autorid soovitavad tüsistusi jagada kergeteks ja rasketeks, kuigi on ka keerulisemaid jaotusi. Kerged tüsistused ei mõjuta tulemust, kuid raskete korrigeerimine vajab operatsiooni või mõjutab oluliselt lõpptulemust.

Mitmetes töödes on analüüsitud jäsemepikenduse ulatust ja leitud, et jäseme pikendamine üle 20–25% on komplitseeritum ja annab halvemaid tulemusi. Uuritud on liigeste funktsiooni pärast jäseme pikendamist ning lihase funktsiooni elektromüograafiliselt ja leitud, et lihas pikeneb, kuid sageli jäävad püsivad kontraktiilsuse häired. Samas on esitatud uurimusi, kus jäseme pikendamise määr on olnud tunduvalt suurem eespool soovitatust ja on saavutatud häid funktsionaalseid tulemusi.

Vaatamata tüsilikkusele on alajäseme pikendamine efekiivne protseduur. Patsientide toimetulek paraneb, invaliidsus väheneb, patsientide psühholoogilis-emotsionaalne seisund paraneb ning enamik neist on saavutatud tulemusega rahul.

Kuna lõplikult ei ole selgitatud endosti-periosti suhet luuregeneratsiooni käigus, olid planeeritud histoloogilised uuringud katseloomadel. Samuti ei ole kirjeldatud kaugtulemuse ja patsiendi rahulolu hindamist pärast jäsemepikenduse pikaaegset jälgimist. See oli põhjendus kliiniliste uuringute läbiviimiseks.

Uurimustöö eesmärgid

1. Uurida luuregeneratsiooni ja selle rakulist koostist küüliku sääreluul pärast kortikotoomiat ja distraktsioonpikendamist (publ I)
2. Uurida luu, periosti ja endosti reaktsiooni roti sääreluul pärast resektsioon-osteotoomiat või perforatsiooni ja periosti isoleerimist (publ II)
3. Uurida reie pikendamise mõju põlveliigese sirutajalihastele (publ III)
4. Hinnata alajäseme traumajärgse lühenemuse ja deformatsioonide korrigeerimise tulemusi kasutades Ilizarovi aparaati (publ IV)
5. Hinnata kaugtulemust ulatuslikul alajäseme pikendamisel Ollier'i tõve korral (publ V)

6. Hinnata alajäseme pikendamise kaugtulemusi ja patsientide rahulolu saavutatud tulemusega (publ IV, V, VI)

Materjal ja meetodid

Uurimuse eksperimentaalses osas on läbi viidud katsed 20 Šinšilla tõugu küülikul. Sääre alumises metafüüsis on tehtud kortikotoomia, säärele on asetatud mini-ringfiksaator nii, et mitte kahjustada luusisest verevarustust. Vastaspoolt on kasutatud kontrolliks. Katsete kestus oli 5–35 päeva.

Järgmises katseseerias on kasutatud 142 Wistar'i rotti, kellest 40-le on tehtud sääreluu ülemise metafüüsi resektsioon ja 68-le metafüüsi 1,5mm perforatsioon. Perforatsiooniga katseloomadest 53 jäeti periost terveks, kuid 15 isoleeriti meditsiinilise luuvahaga periost ja endost. Lõplikuks analüüsiks on kasutatud 18 resektsiooniga ja 25 terve ning 15 isoleeritud periostiga katselooma. Katsete kestus oli 1–42 päeva.

Histoloogiliseks uuringuks on võetud 3 cm sääreluu distaalsest otsast küülikutel ja sääreluu resektsiooniosteotoomia või perforatsiooni piirkond rottidel. Koelõigud on värvitud hematoksüliin-eosiiniga, van Giesoni järgi, altsiaansinisega ja tioniiniga. Elektronmikroskoopiaks kasutatud koetükid on pärast fikseerimist sisestatud vaiku. Mikroskopeerimisel on hinnatud hüpereemiat, luuregeneratsiooni endostaalse ja periostaalse komponendi osakaalu ning luu, kõhr- ja sidekoe vahekorda regeneraadis.

Käesolevas töös on analüüsitud aastatel 1977–1995 53 patsiendil luu meta-epifüsaarses osas tehtud 72 alajäseme pikenduse operatsiooni kaugtulemusi. Kasutatud operatsioonimeetodid olid järgmised: distraktsioonepifüsiolüüs – 13 operatsiooni, luu pikendamine metafüüsist osteotoomiajärgselt – 16 operatsiooni ja kortikotoomiajärgselt 43 operatsiooni. Wassersteini meetodil kahe-etapiline jäsemepikendus luu allotransplantaadi kasutamisega on tehtud 9-l patsiendil 9-l juhul. Keskmine patsientide vanus jäseme pikendamise ajal oli 11,9 aastat. Kortikotoomia tegemiseks on kasutatud omakonstrueeritud osteotoomi, millele on saadud autoritunnistus.

Jäseme pikendamise kaugtulemusi on hinnatud kliiniliselt. Arvutatud on pikendusindeksid ning analüüsitud tüsistused. Seitsmel patsiendil on lisaks kliinilisele uuringule tehtud elektromüograafililised uuringud. On arvutatud reie atroofiaindeks (100–(100×kolme mõõtmise summa opereeritud reiel/kolme mõõtmise summa tervel reiel)). Elektromüograafiliste uuringute andmete alusel arvutati lihase motoorse üksuse rekruteerumine protsentides ja põlveliigese sirutajate väsitatavus.

Patsiendi rahulolu hindamiseks on kasutatud Tjernströmi ja Rehnbergi (1994) poolt väljatöötatud modifitseeritud küsimustikku. Küsimustik on saadetud metafüüsi pikendusega või distraktsioonepifüsiolüüsiga jäseme pikkust korrigeeritud patsientidele, küsimustele vastas 37 patsienti (70%).

Tulemuste analüüs

Histoloogilise uuringu tulemusena on täheldatud, et küülikutel säilus venoosne hüpereemia regeneratsiooni piirkonnas esimese nädala jooksul, kuid siis taandus, mis näitas, et operatsioon ise ei häirinud olulisel määral luu reparatiivseid protsesse. Viiendaks kortikotoomiajärgseks päevaks oli oluliselt tõusnud osteoklastide sisaldus regeneraadis. Luu ja pehme koe vahekorra hindamisel selgus, et opereeritud poolel olid reparatiivsed protsessid tunduvalt kiiremad kui kontrolljäsemes. Resektsiooniosteotoomia piirkond roti sääreluus täitus 28. päevaks luukoega, mille päritolu oli primaarselt periostaalne ja sekundaarselt endostaalne. Roti sääreluu perforatsiooniava täitumise hindamisel selgus, et endostaalne osteogenees on primaarne ja periostaalne on sekundaarne. Endostaalne osteogenees kulgeb praktiliselt ilma kõhrelise faasita ja regenereeruv luu jõuab 28–ks päevaks remodelleerumise faasi. Samas on luukoe endostaalse ja periostaalse kalluse teke pidurdunud, kui periost on endostist isoleeritud.

Elektromüograafilise uuringu tulemusel selgus, et kõigil 7-l reiepikendusega haigel oli tekkinud reielihaste atroofia (atroofiaindeks 1,9 – 5,8%). *M.vastus medialise, m. rectus femorise* ja *m. vastus lateralise* rekruteerumine on opereeritud jäseme lihastel võrreldes terve poolega vähenenud 30–50% ning samas on väsitatavus samapalju suurenenud. Lihase rekruteerumist ja väsitatavust mõjutas jäsemepikenduse ulatus, kuid jäsemelühenemise põhjus (omandatud või kaasasündinud), jäsemepikenduse meetod, kiirus ja pikendamise kestus neid parameetreid ei mõjutanud.

Traumajärgsel jäseme pikendamisel on analüüsitud 14 patsiendi jäsemepikendust neljas erinevas grupis. Keskmine pikendusindeks eri gruppides oli 20,5 – 33,5 päeva sentimeetri kohta. Ühel patsiendil on reie pikendust tehtud kolmel korral viieaastase vaheajaga, iga pikendus 5 cm. Kolmandal reie pikendusel oli pikendusindeks 46,4 päeva sentimeetri kohta, mis viitab sellele, et korduv jäseme pikendamine vähendab luu repratiivseid võimeid. Tüsistusi oli 28% patsientidest.

Ollier'i tõvega patsiendil, kellel oli väljendunud jäseme lühenemus – 32 cm ja reieluu ning põlveliigese deformatsioon, on tehtud reie ja sääre pikendus vastavalt 22 ja 10 cm. Histoloogiline uuring näitas, et embrüonaalsed osteokondraalsed Ollier'i tõvele iseloomulikud kolded luus olid pikenduse käigus luustunud. Patsient on jäsemepikenduse funktsionaalse ja kosmeetilise kaugtulemusega (jälgimisaeg 25 aastat) väga rahul.

Patsiendi rahulolu küsimustikule vastanute protsent oli 70%, jälgimise aeg oli keskmiselt 19,4 (10–29) aastat, suuremal osal üle 15 aasta. Pikenduseindeks oli keskmiselt 29,5 päeva/cm, seejuures 33,6 kaasasündinud ja 23,7 osteomüeliidijärgsel jäseme lühenemisel. Nende gruppide vaheline erinevus oli statistiliselt tõene. Tüsistuste arv oli mõõdukas ning esines kokku 49% patsientidest, kuid raskeid ja kergeid tüsistusi vastavalt 21% ja 28%. Patsiendid olid, vaatamata asjaolule, et 65% neist lonkab sageli või pidevalt, suures osas rahul saavutatud üldise (78%) ja kosmeetilise tulemusega (68%) ning 81% neist laseks operatsiooni teha uuesti, kui see peaks vajalik olema.

Uurimistööst tulenevad järeldused

1. Eksperimentaalset luuregeneratsiooni kortikotoomia ja distraktsioonpikenduse järel iseloomustas küülikul varane osteoklastiline resorptsioon ja osteoblastiline proliferatsioon. Regeneratsioon pikendataval jäsemel oli kiirem kui sääreluu enkondraalne ossifikatsioon kontrolljäsemel. Luuregeneraadi verevarustus ei olnud häiritud, sest venoosne hüpereemia esines ühe nädala jooksul pärast operatsiooni.
2. Sääreluu defekti täitumine resektsiooniosteotoomia järel oli rotil sarnane embrüohistogeneesiga primaarse periostaalse ja sekundaarse endostaalse ossifikatsiooniga. Luu kortikaalkihi perforatsiooni järgselt toimus endostaalne ossifikatsioon ilma kõhrelise staadiumita, aga periostaalne ossifikatsioon üle kõhrelise staadiumi. Isoleeritud periosti korral oli endostaalse kalluse teke pidurdunud ja periostaalse kalluse teke enamasti puudus.
3. Jäseme pikendamine reiest kahjustas põlve sirutajalihaseid (motoorsed üksused), seejuures sõltus kahjustus jäsemepikenduse ulatusest ja ei sõltunud jäseme lühenemise põhjusest, jäsemepikenduse meetodist, pikendamise kiirusest ja kestusest. *M.vastus medialise* kahjustus oli suurem kui *m.rectus femorisel* ja *m.vastus lateralisel.*
4. Epifüsaarplaadi enneaegse posttraumaatilise sulgumise ja suure luudefekti korral kasutati nelja erinevat jäsemepikendamise meetodit, sõltuvalt kahjustuse iseloomust. Luuregeneraadi ossifikatsioon ei olnud häiritud ja tüsistuste sagedus oli väike. Korduvad jäsemepikendused võivad mõjutada luuregeneratsiooni.
5. Ollier'i tõve haigel oli ulatuslik jäseme segmendi pikendus (75% reie algpikkusest) läbi osteokondraalsete kollete. Pikenduse käigus täitusid embrüonaalse kõhrega osteokondraalsed kolded luukoega. Saavutati 32 cm-ne jäsemepikendus, tüsistusi ei esinenud. Patsient oli 25 aastase jälgimisperioodi järel ravitulemusega rahul.
6. Jäseme distraktsioonpikendus epimetafüsaarses osas tehti 53-l patsiendil, seejuures tehti enamasti kortikotoomia (57%) või osteotoomia (26%) spetsiaalse osteotoomiga. Saavutati keskmiselt 26,2% (8,5–81%) jäseme pikendust, keskmine pikendusindeks oli 29,5 (12,9–59,4) päeva ühe cm kohta.
7. Kaugtulemuste (keskmiselt 19,4 aastat) analüüs näitas, et 78% patsientidest olid rahul saavutatud tulemusega ja 68% kosmeetilise efektiga, kusjuures see sõltus tüsistuste esinemisest ja patsiendi soost. Kuigi 81% patsientidest olid nõus jäsemepikendamise kordamiseks, kui see osutuks vajalikuks, ei seostunud see otsus objektiivselt hinnatud funktsionaalsete näitajatega.

Käesolevas töös on eksperimentaalselt ja kliiniliselt uuritud alajäseme pikendamist. Eksperimentaalsed uuringud näitasid endostaalse luuregeneratsiooni tähtsust ja selle koosmõju periostaalse ossifikatsiooniga. Edasised uuringud on vajalikud, et selgitada põhjalikumalt luuregeneratsiooni rakulist ja molekulaarset koostist. Kliinilistel uuringutel selgus, et erinevate operatsioonimeetodite vahel ei ole olulist erinevust, kuid tulemused erinesid etioloogilistes gruppides. Patsiendid olid rahul kaugtulemusega. Kuigi praegusel ajal on laialdast kasutamist leidnud jäsemepikendamine intramedullaarsel naelal, jäävad uuritud meetodid liigeselähedaste deformatsioonide korral kasutusse. Seetõttu on vajalik jätkata uuringuid operatsioonimeetodite täiustamiseks. Kuna varasemad pikaaegsed kaugtulemuse uuringud puuduvad, siis on vajalik uuringuid ka selles valdkonnas jätkata.

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PUBLICATIONS

A. Märtson, P. Roosaar. A morphological research of the bone regeneration in leg lengthening. (Experimental study). Topical problems of treatment skeletal injuries and diseases. Acta et Commentationes Universitatis Tartuensis 835. Tartu, 1989. p.79–88. (rus)

P. Hussar, A. Piirsoo, A. Märtson, A. Toom, T. Haviko, Ü. Hussar – Bone healing models in rat tibia after different injuries. Annales Chirurgiae et Gynaecologiae 2001; 90:271–9

Ü. Kaljumäe, A. Märtson, T. Haviko, O. Hänninen. The effect of lengthening of the femur on the extensors of the knee. J. Bone Joint Surg 1995; 72A(2): 247–50

Märtson, T. Haviko. Posttraumatic limb shortening: treatment with the Ilizarov distraction device. Annales Chirurgiae et Gynecologiae 2000; 89: 303–7

Märtson, T. Haviko, K. Kirjanen. Extensive limb lengthening in Ollier's disease: 25-year follow-up. Medicina 2005; 41(10): 861–6

Märtson, K. Kirjanen, A. Toom, T. Haviko. Outcome and satisfaction studies in limb lengthening. International Orthopaedics. Submitted Int Orthopaedics

OUTCOME AND SATISFACTION STUDIES IN LIMB LENGTHENING

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ABSTRACT

In this study 72 limb lengthening operations performed on 63 lower limbs in 53 patients (22 male and 31 female) were analysed. Distraction epiphysiolysis, metaphyseal osteotomy and corticotomy were performed using the Ilizarov device. The mean age of the patients at the time of operation was 11.9 years. A database was created of the stored follow-up data of the patients and of the data of the questionnaire designed for the present study. Thirty-seven patients (70%) responded, mean follow-up being 19.4 years (range 10–29). The t-test for paired or unpaired data and regression analysis for comparison between the groups to assess the influencing factors were used. Different methods of limb lengthening do not differ significantly in long-term results. Of the patients 78% were satisfied with the present situation and 68% were satisfied with the cosmetic appearance. Although the complication rate of limb lengthening was not high (49%), no one of complications affect final result and 81% of the patients agree to pass the procedure once more.

INTRODUCTION

More active osteogenesis and greater mechanical strength have been described as the two advantages to metaphyseal lengthening [15]. Treatment of limb length inequality is a complicated and long-term procedure and one must carefully equalize limb length in a manner that is neither physically nor emotionally scarring [9]. Brockway and Fowler stated (1942) already, “Leg lengthening will always entail certain hazards and there will probably be some poor results, but with the experience gained we feel that the hazards and poor results can be reduced to a creditable minimum.” They gained good or satisfactory results in 87% of limb lengthening patients.

Psychological and social impact studies of the Ilizarov leg lengthening procedure concluded that nearly all patients and parents had positive attitude to the procedure and the support that they received. Most patients, 88%, stated that they would undergo the treatment again, if it was indicated [18]. According to another study, 82% of patients were satisfied with the over-all result, but only 67% of children thought that the duration of the treatment was reasonable and said that they would be willing to have the procedure a second time if it was needed [8]. Sixteen of 22 patients completed a questionnaire after leg lengthening according to Wagner. Most noted both improved function and cosmetic appearance. The answer to the question “Would you do it again?” was yes in 11 and no in 5 cases. Eight of the 22 patients did no experience any psychological problem [10]. In a study of Tjernström and Rehnberg (1994) 60 patients were satisfied with the results of lengthening. Only five patients, all with traumatic shortenings, were dissatisfied. Complete patient satisfaction was achieved in 45 of 48 patients (94%) and partial satisfaction was achieved in the remaining three [17].

The aim of the study was to investigate the clinical outcome, patient satisfaction and long-term results of lower limb lengthening. The study was designed as a retrospective and case control study with a statistical analysis of the database and questionnaire data.

MATERIAL AND METHODS

In this study 72 limb lengthening operations performed on 63 lower limbs in 53 patients (22 male and 31 female) were analysed. The mean age of the patients at the time of operation was 11.9 years.

Limb length discrepancy was measured clinically and by plain x-ray. The indications for limb lengthening were lower limb shortening more than 3 cm or a severe deformity at the joint level.

The Ilizarov external device was used. All operations were performed in general anaesthesia.

For detection of the metaphysis, the AO quadrate method was used. In cases of corticotomy the bone was not broken to prevent damage of the cancellous bone and the vessels inside it. For this purpose in our clinic, a special crescent-shaped osteotome was designed consisting of a handle and a cutting edge covered with a plate which protects soft tissues and prevents incision of the spongious bone (Invention No1297826 USSR).

The causes of limb shortening were: congenital 27 (51%), achondroplasia 6 (11 %), posttraumatic 8 (15%), sequelae of epiphyseal osteomyelitis 8 (15%), and other 4 (8%) cases. The operations (72) performed for limb lengthening were metaphyseal corticotomy, 43 cases (60%), metaphyseal osteotomy, 16 cases (22%), and distraction epiphysiolysis, 13 cases (18%). In 8 cases of tibial lengthening preliminary Achilles tendon lengthening by Z-plasty was performed.

Distraction began on the fifth day at a rate of 1 mm per day in four equal doses applied.

The data collected from follow-up were: age, shortening, diagnosis, surgery, lengthening, function, complications. Data analysis was performed in two stages: analysis of the follow-up data and analysis of the questionnaire data.

Minor and major complications were registered. Minor complications were transient swelling and infection surrounding the pin; major complications were those that required additional treatment or affected the final result.

Physical treatment and dynamization was applied in the case of every patient, partial to full weight bearing was allowed as tolerated with the external fixator in place.

The percentage of lengthening (length of the distraction gap divided by the initial length of the bone segment multiplied by 100%) and lengthening indices (days needed for lengthening/centimetres of lengthening) were calculated.

A questionnaire developed originally by Tjernström and Rehnberg (1994) modified for this specific study was used (Table 1).

Table 1. A questionnaire for specific study.

Follow-up questionnaire				
I Case				
Age				
Sex				
employed	studying	unemployed	disabled	home bound
II Complaints				
1. Back pain:				
Pain		Appearance		Localization
absent		none of the time		thoracic
a little		a little of the time		lumbar
moderate		some of the time		total
serious		most of the time		
extreme		all of the time		
2. Hip pain:				
Pain		Appearance		Localization
absent		none of the time		right hip
a little		a little of the time		left hip
moderate		some of the time		both
serious		most of the time		
extreme		all of the time		
3. Knee pain:				
Pain		Appearance		Localization
absent		none of the time		right knee
a little		a little of the time		left knee
moderate		some of the time		both
serious		most of the time		
extreme		all of the time		

4. Limping	Appearance
	1. none of the time
	2. a little of the time
	3. some of the time
	4. most of the time
	5. all of the time
III	
1. Do you remember what cumbered you before limb lengthening?	
2. Do you remember how you compensated for your limb length? (scoliosis, pelvic tilt, flexed knee, equinus, heel lift, supporting brace)	
3. Did you have pain before limb lengthening?	
4. Did you have pain during limb lengthening?	
5. Are you satisfied with the present situation?	
	1. Fully satisfied
	2. Partly satisfied
	3. Not at all satisfied
6. Are you satisfied with the present cosmetic situation?	
	1. Fully satisfied
	2. Partly satisfied
	3. Not at all satisfied
7. Do you wear a heel lift? Yes No	
8. If you needed to pass the limb lengthening once more, would you agree?	
	1. Yes
	2. No Why?

Where it was appropriate, t-test was employed for paired or unpaired data.

In order to obtain clear and comparable data about pain and subjective satisfaction on the basis of the follow-up questionnaire, the following scoring was used:

1) Pain intensity was scored so that "absent" was marked as "1" and "extreme", as "5".
2) Appearance of pain was scored so that "none of the time" was marked as "1" and "all of the time", as "5".
3) Satisfaction was scored so that "fully satisfied" was marked as "1" and "not at all satisfied", as "3".
4) Limping was scored so that "non of the time" was marked as "1" and "all of the time", as "5".

These scores were used in calculations with continuous data and in t-statistics for the patient groups. For the distributions with a larger patient number and lesser divisions in the overall study group, chi-square test was applied. In both cases a p-value less than 0.05 was considered statistically significant. Regression analysis was performed for comparison between the groups to assess the affecting factors.

RESULTS

A database was created of the stored follow-up data of the patients and of the questionnaire data. Altogether 63 lower limbs were lengthened in 53 patients (total 72 operations).

The amount of lengthening of one limb segment was 1–22 cm. In minor cases mostly angular deformity correction was done. There were altogether 26 (in 49% of patients) complications, among which 15 (28%) were minor in and 11 (21%) were major complications.

A mean 26.2% (range 8.5–81) of lower limb lengthening was gained. The percentage of limb lengthening differed significantly in achondroplasia cases, 65.3±7.9%, versus congenital cases, 16.0±0.8% ($p<0.0001$), and versus sequelae of osteomyelitis cases, 20.2±2.8% ($p=0.0029$). Mean lengthening index was 29.5 (range 12.9–59.4) days per centimetre but it varied between the study groups (Table 2).

Table 2. Lengthening index depending on diagnosis.

Diagnosis (number of patients)	Lengthening percentage	Lengthening index (days per centimetre)	Significance
Achondroplasia (6)	65.3±7.9	22.2±2.0	*
Congenital shortening (27)	16.0±0.8	33.6±2.3	*, **
Sequelae of osteomyelitis (8)	20.2±2.8	23.7±1.9	**
Posttraumatic shortening (8)	13.5±1.1	30.4±2.4	
Others (4)	32.6±14.7	33.4±7.5	

* – $p=0.0088$; ** – $p=0.0245$

A questionnaire was sent to 53 patients. Response rate was 70% (37 patients); 15 men and 22 women responded. Four patients with achondroplasia, 17 with congenital shortening, 6 with sequelae of osteomyelitis, 7 with posttraumatic shortening and 3 patients with other diagnoses responded.

Mean follow-up time was 19.4 years (range 10–29). Only one of the examined patients has disability, three of them (all women) are at home nursing children and the rest are employed or attend school. The majority, 26 patients (70%), remember that something cumbered them before the surgery. The reasons for cumbering were: difference in the length of the lower legs in 15 patients, limping in 5 patients, short stature in 3 patients and other in 3 patients. For compensation for the deformity, more than a half of the patients 54% (20 patients), used walking aids or a shoe lift. Despite the fact that 24 (65%) of the respondents are limping most of the time or all of the time, 29 (78%) are satisfied with the present situation and 25 (68%) are satisfied with the cosmetic appearance. If there arose the need to pass limb lengthening once more, 30 (81%) of the examined patients would agree to do it. Although 81% of the patients agreed to pass limb lengthening once more if needed, this decision was not correlated with any objectively estimated functional indicator one year postoperatively.

As of the 37 patients who responded 15 had complications, the remaining 11 complications occurred in 16 patients who did not respond. Satisfaction depended on the complications and gender (Table 3).

Table 3. Chi-square analysis of patients satisfaction with limb lengthening.

Satisfaction	Complications/gender	Satisfied	Non satisfied	Significance
Present situation	Yes	8	7	p<0.000001
	No	21	1	
Cosmetic effect	Yes	7	8	p=0.000011
	No	18	4	
Readiness to pass limb lengthening once more	Yes	11	4	p=0.0138
	No	19	3	
Scars	Men	11	4	p=0.0069
	Women	8	14	
Cosmetic effect	Men	13	2	p=0.0109
	Women	12	10	

After the limb lengthening the function of the knee joint was impaired as expressed in the diminishing of knee flexion by a mean of 79.1º±5.3º, which differed significantly from the preoperative values (p<0.0001). The degree of the impaired function of the knee did not significantly differ for the method of operation used. After one year the function was significantly improved, but flexion was still 12.3º±4.0º less than the preoperative values (p=0.0103). The patients in the distraction epiphysiolysis group gained almost the same level of knee flexion after one year, 143º±2.1º, versus preoperative 145º±1.7º (p=0.4697). The patients with metaphyseal osteotomy showed a larger difference between the preoperative (128.2º±7.6º) and one-year postoperative (115.0º±7.6º) knee flexion values, but there was no statistical difference (p=0.2289). A statistically significant difference in knee flexion was found in the metaphyseal corticotomy patients: the preoperative value being 136.5º±2.6º and the one-year postoperative value being 121.7º±5.6º (p=0.0186).

The appearance of pain and the pain intensity in the knee were in the negative correlation with knee flexion and with the change in knee flexion. It is obvious that if there is pain in the knee joint its function is more impaired.

Mainly low back pain was observed in 15 patients, but it was considered serious only in two patients. Hip pain was noted in 11 patients and it was serious or extreme in 2 cases. Knee pain was observed in 18 patients and it was serious or extreme in 3 cases. At the same time, 13 patients stated that they did not have any pain in any segment of the limb or in the back. Among them 2 were achondroplasia patients, 4 were congenital shortening, 2 were sequelae of osteomyelitis and 4 were posttraumatic shortening patients.

The score of the intensity of back pain for the patients who underwent distraction epiphysiolysis was 2.50±0.62 (on the border between “a little” and “moderate”), 1.40±0.15 for the corticotomy patients (on the border between “absent” and “a little”) and 1.73±0.30 for the osteotomy patients (on the border between “absent” and “a little”). A statistically significant correlation occurred between the distraction epiphysiolysis group and the corticotomy group (p=0.0168).

Satisfaction with the present situation differed in the context of the method of operation (Fig.1). Difference in satisfaction with the present situation between the genders was absent. Some patients who were generally satisfied with the cosmetic result were not satisfied with the appearance of scars (Table 3).

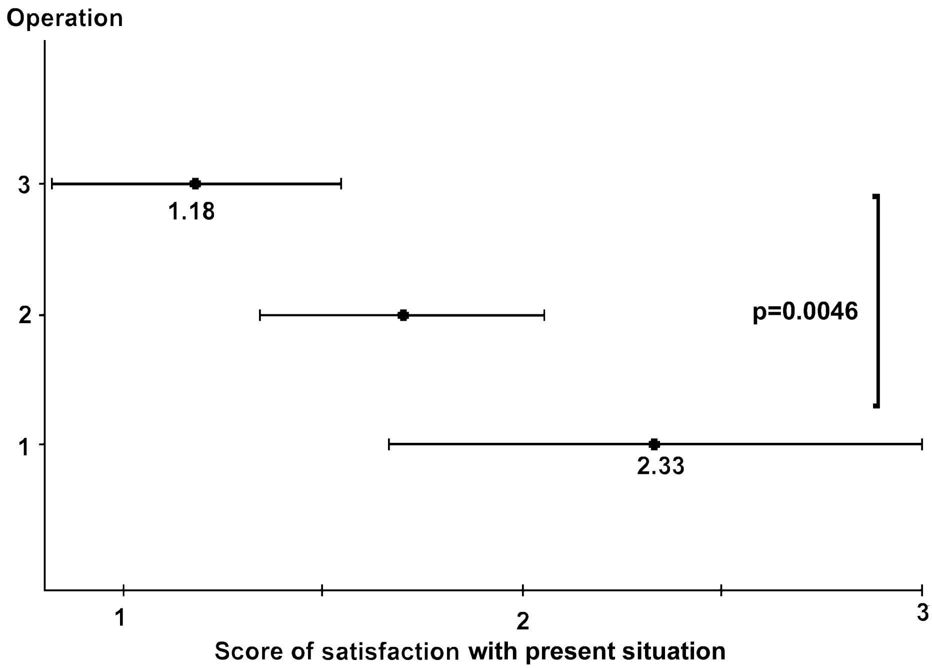

Figure 1. Score of satisfaction with present situation. 1 – distraction epiphysiolysis; 2 – metaphyseal corticotomy; 3 – metaphyseal osteotomy. Bars indicate standard deviation from the mean score.

Among the 17 patients who had used a heel lift or a brace and had had some compensative deformity before limb lengthening, only one would not have agreed to pass limb lengthening operation once more. Among the 20 patients who did not use any aid before the surgery, six would not have agreed to pass the surgery once more (p=0.035).

For the patients who would agree to pass limb lengthening once more the score of hip pain in the present study was 1.37±0.15 (on the border between "absent" and "a little") and for those who would not agree to undergo this procedure once more the corresponding score was 2.72±0.52 (on the border between "a little" and "moderate") (p=0.0015). Similar difference was observed in the score of appearance of hip pain between those who would agree (1.47±0.18; on the border between "none of the time" and "a little of the time") and those who would not agree to pass the surgery once more (2.43±0.37; on the border between a little of the time and some of the time) (p=0.028). At the same time it should be mentioned that neither pain intensity nor appearance of pain in the hip affected satisfaction with the present situation.

DISCUSSION

In the present study 53 patients with metaphyseal lengthening and distraction epiphysiolysis were analysed. For performing corticotomy, we used a special curved osteotome invented by us; a similar instrument have been employed by other researchers as well [12]. Metaphyseal lengthening was preferred to diaphyseal lengthening since metaphyseal bone is expected to heal faster than diaphyseal bone [5]. White and Kenwright (1991) concluded that the level of the osteotomy is located at the junction between the metaphysis and the diaphysis, an area with a greater potential for bone healing than the mid-diaphysis.

The lengthening indices were calculated, the average for the present study being 29.5 days per centimetre. Lengthening index is used for the description of bone lengthening [14]. Calculation of the time required for the lengthening of a lower limb segment by one percent [16] did not seem necessary, as in comparison of the lengthening indices for groups with different aetiology, the techniques used, and the site of the procedure gives enough valuable information about a limb lengthening procedure [1, 2, 4, 6, 11, 17, 22, 23]. Lengthening indices are larger in congenital cases because of the influence bone reparative potency. In postinfection cases we did not observe elongation of bone regeneration time, however this can be explained by very young patient age in epipyseal osteomyelitis cases and probably also by the fact that infection did not affect bone regeneration potency.

Sometimes good results may be obtained in extensive limb lengthening, e.g. in achondroplasia. The lengthening indices and the percentages of limb lengthening differ significantly between congenital and achondroplasia groups and between the postinfection and achondroplasia groups. Short stature and very short limbs, as well as the performance of operations at double sites in achondroplasia cases usually explain this difference. It can be concluded that in most – 37 – limbs the lengthening index in the present study was below 30 days per centimetre.

The amount of lengthening at an acceptable complication rate should not exceed 25% [13]. Actually, the lengthening gained in most of the patients remained lower than 20%, which may explain quite good results in the present study. Complication rate has varied with respect to the amount and method of limb lengthening, ranging from 30% [7] to even 1.2 major complications per patient [9]. Complication rate reached 49% and was not high in the present study. No one complication affected the final result in our study. At the same time, it seems impossible to avoid minor complications when external fixators are used [21].

Mean follow-up time for the patients was 19.4 years. Response rate was high (70%). Yet those who did not respond (11 complications in 16 patients) did not do so because of the poor result only but for other reasons as well. At least five of such patients are living abroad and the questionnaire did not reach to them.

Despite the fact that 24 (65%) of the responded patients are limping most or all of the time, 29 (78%) are satisfied with the present situation and 25 (68%) are satisfied with the cosmetic appearance. High satisfaction is demonstrated by the fact that 30 (81%) of the patients would pass limb lengthening once more if it were indicated. However, as the question "Would you do it again?" is very emotional, most patients agree to pass the surgery once more.

Although the pain scores for different locations were calculated, we did not observe significant statistical correlation between diagnosis, method of operation used and other indicators. Only pain in the knee joint is associated with more impaired function.

Already Brockway and Fowler (1942) had good or satisfactory results in 87% of limb lengthening patients. Most patients, 88%, stated that they would undergo the treatment again if it were indicated [18]. According to another study, 82% of patients were satisfied with the overall result, but only 67% of children thought that the duration of the treatment was reasonable and said that they would be willing to have the procedure a second time if it were indicated [8]. Patients were also more satisfied with thigh scars than with leg scars because clothing can easily cover thigh scars [11]. In the present study, some patients who were generally satisfied with the cosmetic result were not satisfied with the appearance of scars.

There is no significant difference between the methods used – distraction epiphysiolysis showed even better functional outcome despite the fact that the procedure was performed close to the joint. Yet these patients were more dissatisfied with the result than the others. There is still room for limb lengthening from the epimetaphyseal region of the bone, especially when there is a need for a simultaneous deformity correction.

Limb lengthening requires careful planning and meticulous outpatient care and is still burdened by minor complications. Experience may lessen major complications, however, pin tract problems, swelling, and pain will plague these patients. A large group of patients can benefit from lengthening procedures, and new techniques are improving the risk-benefit balance. A thoughtful approach is needed by the surgeon, especially taking time to fully inform the patient about the time, effort, and problems involved in substantial limb lengthening.

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REFERENCES

1. Aronson J. Limb lengthening, skeletal reconstruction and bone transport with the Ilizarov method. J Bone Joint Surg 1997; 79A; 1243–58
2. Bonnard C, Favard L, Sollogub I, Glorion B. Limb lengthening in children using the Ilizarov method. Clin Orthop. 1993; 293: 83–8
3. Brockway A, Fowler SB: Experiences with 105 leg-lengthening operations. Surg Gynecol Obstet 1942; 72:252–6
4. Eralp L, Kocaoglu M, Ozkan K, Turker M. A comparison of two osteotomy techniques for tibial lengthening. Arch Orthop Trauma Surg 2004; 124(5): 298–300
5. Fischgrund J, Paley D, Suter C. Variables affecting time to bone healing during limb lengthening. Clin Orthop 1994; 301: 31–7
6. Franke J, Hein G, Simon M, Hauch S. Comparison of distraction epiphyseolysis and partial metaphyseal corticotomy in leg lengthening. Intl Orthop 1990; 14: 405–13
7. Garcia-Cimbrelo E, Olsen B, Ruiz-Yague M, Fernandez-Baillo N, Munuera-Martinez L. Ilizarov technique. Results and difficulties. Clin Orthop 1992; 283: 116–23
8. Ghoneem H, Wright J, Cole WG, Rang M. The Ilizarov method for correction of complex deformities. Psychological and functional outcomes. 1996; 78A(10): 1480–5
9. Guidera KJ, Hess WF, Highhouse KP, Ogden JA. Extremity lengthening: results and complications with the Ortofix system. J Pediatr Orthop 1991; 11(1): 90–4
10. Hrutkay JM, Eilert RE. Operative lengthening of the lower extremity and associated psychological aspects: the Children's Hospital experience. J Pediatr Orthop 1990; 10(3): 373–7
11. Karlen LKP, Yinusa W, Yan LS, Wang KW, Hoi LY, John LCY. Analysis of scar formation after lower limb lengthening – Influence on cosmesis and patient satisfaction. J Pediatr Orthop 2004; 24(6): 706–10
12. Kristiansen PL, Steen H. Reduced lengthening index by use of bifocal osteotomy in the tibia: comparison of monofocal and bifocal procedures with the Ilizarov external fixator. Acta Orthop Scand 2002; 73: 93–7

13. Maffuli N, Fixsen JA. Distraction osteogenesis in congenital limb length discrepancy: a review. Journal of the Royal College of Surgeons of Edinburgh. 1996; 41(4): 258–64
14. Monticelli G, Spinelli R. Leg lengthening by closed metaphyseal corticotomy. Ital J Orthop Traumatol 1983; 9(2): 139–50
15. Moseley CF. Leg lengthening. A review of 30 years. Clin Orthop 1989; 247: 38–43uj
16. Noonan K, Leyes M, Forriol F, Canadell J. Distraction osteogenesis of the lower extremity with use of monolateral external fixator. A study of two hundred and sixty one femora and tibiae. J Bone Joint Surg 1998; 80A: 793–806
17. Paley D. Problems, obstacles, and complications of limb lengthening by the Ilizarov technique. Clin Orthop 1990; 250: 81–104
18. Ramaker RR, Lagro SWJ, van Roermund PM, Sinnema G. The psychological and social functioning of 14 children and 12 adolescents after Ilizarov leg lengthening. Acta Orthop Scand 2000; 71(1): 55–9
19. Tjernström B, Rehnberg L. Back pain and arthralgia before and after leg lengthening. Acta Orthop Scand 1994; 65(3): 328–32
20. White SH, Kenwright J. The importance of delay in distraction of osteotomies. Orthop Clin North Am 1991; 22(4): 569–79
21. Yeu K, Huang SC, Liu TK. Treatment of malunion of the lower extremities by the Ilizarov technique. Formos Med Assoc 1994; 93: 403–10
22. Yun AG. Severino R. Reinker K. Attempted limb lengthenings beyond twenty percent of the initial bone length: results and complications. J Pediatr Orthop 2000; 20(2): 151–9
23. Zarzycki D, Tesiorowski M, Zarzycka M, Kacki W, Jasiewicz B. Long-term results of lower limb lengthening by physeal distraction. J Pediatr Orthop 2002; 22(3): 367–70

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