Case Report
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Accordion maneuver with Ilizarov placed plate *in situ*, in a case of femoral shaft non-union and re-fracture with quiescent infection, discussion on treatment strategies: a case report

Abstract

Introduction: Aim of this study to evaluate the clinical efficacy and highlight their relevance in present orthopedics practice where multiple newer choices have been become famous. Accordion maneuver design with modified Ilizarov apparatus, a single stage procedure in femoral shaft non-union and re-fracture with quiescent infection place dynamic compression plate (DCP) *in situ*. Femoral shaft fractures (FSFs) are frequently occurring injury due to RTA. Infection (0.4%) and non-union (1.1 - 14%) are the morbid complications of femur fractures. The healing rate for femora shaft non-unions is too high (90%). [1] Accordion Maneuver (AM) is the "Bloodless Stimulation" of bone healing described by Professor G. A. Ilizarov. It comprises of alternate compression and distraction which produce stress in living tissue and also convert biologically inactive scar tissue at non-union site into tissue capable of neo-osteogenesis.[2] The suggested treatment for quiescent type of infected non-union is a single stage procedure with minimal or no debridement and if implant is provide sufficient stability it placed *in situ*.[3]

Case Report: In the case, a 26 years man treated by DCP with MIPO for his Gustilo type – I open, comminuted fracture of mid femoral shaft (Left) following RTA. Post-operative infection occurred within 2 weeks of operation. Infection controlled by exploration and surgical toileting with antibiotic. After four (4) months later he was sustained re-fractured with bending plate due to fall again. Finally he was diagnosed as H. Rosen`s type 3 Quiescent infected non-union (Oligotrophic) and Romano stage -1 post-implant infection with re-fracture mid shaft of left femur. Then he was treated following accordion principles with modified Ilizarov frame. AM was applied according to protocol of Baruah and Patowary of non-union treatment.

Result: After complete union and consolidation, substantiated by radiological evidence Ilizarov apparatus was dispelled six (6) months later of installation without removal of plate and four (4) months of that finally plate was removed. After one (1) year and four (4) months of mounted Ilizarov frame, the patient was in full free movement of knee and hip. He had no problem during walking even running.

Conclusion: We pursue for the treatment, accordion maneuver with Ilizarov apparatus, a single stage procedure in femoral shaft non-union and re-fracture with quiescent infection kept plate *in situ*. Few authors reported, Accordion Maneuver (AM) techniques with Ilizarov apply over intramedullary nail (IMN) *in situ* for aseptic non-union of femur. In this study, we discussed the role of this tool (AM) for the treatment of femoral shaft non-union and re-fracture with quiescent infection place plate (DCP) *in situ*.

Message of the case report: This case become an empirical innovation, due to after a thoroughgoing literature review, we are not bringing out any similar case. In this study we discuss briefly about AM at mechanical and cellular level. Kept implant *in situ* provides stability and preserve soft tissue, vascularity and bony fragment in position. AM with Ilizarov apparatus deliver best outcomes in infected non-union management.

Keywords: accordion maneuver, compression, distraction, distraction osteogenesis, femoral shaft fracture, infected non-union, quiescent infection, re-fracture

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Abbreviations: AM, accordion maneuver; CBC, complete/ full blood counts; C/S, culture and sensitivity test; CRP, c-reactive protein; DO, distraction osteogenesis; ESR, erythrocyte sedimentation rate; EMPs, extracellular matrix proteins; FIS, fixation *in situ*; FSFs, femoral shaft fractures; PF, postponed/ neglected fracture; POD, post-operative day; SSI, surgical site infection; SSSI, superficial surgical site infection

Introduction

Aim of this study to evaluate the clinical efficacy of accordion maneuver design with Ilizarov apparatus, a single stage procedure in femoral shaft non-union and re-fracture with quiescent infection place dynamic compression plate (DCP) *in situ*.

Femoral shaft fractures (FSFs) are frequently occurring injury due to RTA, which are 10-21 per 100,000 in a year.[4] Infection and non-

union are the morbid complications of femur fractures– infection rate 0.4 and non-union 1.1 to 14%.[5,6] There are several treatment options in fracture shaft of femur but no standard guidelines. The gold standard treatment for femoral shaft fracture is intramedullary nailing (IMN). Now a days DCP with MIPO technique also popular fixation method for comminuted fracture. Re-fractures (6.5-14.2%) are another complication by further trauma causes implant failure in the region of improper callus formation.[7]

In the case, a 26 years man treated by DCP with MIPO for his Gustilo type-I open, comminuted fracture of mid femoral shaft (Left) following RTA. Post-operative infection occurred within 2 weeks of operation. Infection controlled by exploration and surgical toileting with antibiotic. He was re-fractured with bending plate after fall again. Then he was categorized as diagnosed of H. Rosen`s type 3 Quiescent infected non-union (Oligotrophic) and Romano stage-1 post-implant infection with re-fracture mid shaft of left femur.

Accordion Maneuver (AM) is the "Bloodless Stimulation" of bone healing described by Professor G. A. Ilizarov. It comprises of alternate compression and distraction which produce stress in living tissue and also convert biologically inactive scar tissue at non-union site into tissue capable of neo-osteogenesis.[2] The suggested treatment for quiescent type of infected non-union is a single stage procedure with minimal or no debridement and if implant is provide sufficient stability it placed *in situ*.[3] The healing rate for femora shaft non-unions is too high (90%).[1]

We pursue for the treatment, accordion maneuver with Ilizarov apparatus, a single stage procedure in femoral shaft non-union and re-fracture with quiescent infection kept plate *in situ*.

We are not been susceptible to trace any publication indicating treatment of quiescent type infected non-union of femoral shaft fracture or others bone fracture, over plate *in situ* by modified Ilizarov frame with accordion maneuver (AM). Circular external fixators using Ilizarov`s principles give consistent outcomes in such difficult scenarios, disparate others conventional procedure. It stimulate and initiate neo-osteogenesis without bone grafting and also regenerate soft tissue beside plastic reconstruction surgery.[8]

Few authors reported, accordion maneuver (AM) techniques with Ilizarov apply over intramedullary nail (IMN) *in situ* for aseptic non-union of femur. In this study, we discussed the role of this tool while treating femoral shaft non-union and re-fracture with quiescent infection place dynamic compression plate (DCP) *in situ*.

After a thoroughgoing literature review, we are not bringing out any similar case, which mark our case as an empirical innovation. Our aim is to assess the results of accordion maneuver (AM) with modified Ilizarov osteosynthesis in such case and highlight their relevance in present orthopedics practice where multiple newer choices have been become effective and famous.

Case report

A 26 years young male fall on road traffic accident 4 years back. After that high energy trauma he was experienced pain, bleeding and unable to move of left lower limb at thigh region. He was diagnosed a case of open Gustilo type-I, comminuted fracture of mid femoral shaft (Left). Primary resuscitation, wound dressing was done and apply skin surface traction by Thomas Splint for temporary immobilization.

All essential investigations were done and found within normal range. Three (3) days after trauma the fracture was fixed by Dynamic Compression Plate (DCP) with Minimally Invasive Plate Osteo-synthesis (MIPO) procedure. Post-operative radiograph ensured bony fragments and plate ware in potions with maintaining well alignment with minimal fracture gap between fragments (Figure 1).

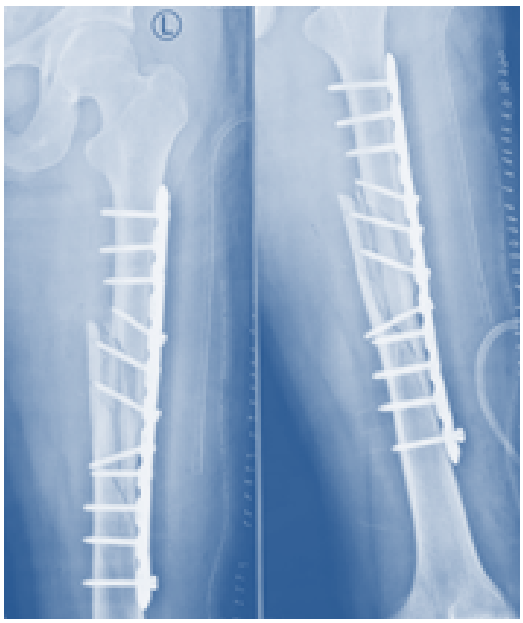

Figure 1 Post-operative x-ray after Plate Osteo-synthesis.

On 14th POD superficial infection was evident. Exploration and surgical toileting was done on same day, wound dressing done daily. Subsequent follow up was going on in improvement of the patient (Figure 2). After 4 months of operation, he was fall down and affected fractured limb (Left lower limb). During physical examination we found moderate tenderness without any abnormal mobility at fractured site. There were no signs of infection. Knee and hip joints movement restricted due to pain. In radiograph there was re-fracture of mid shaft of left femur with plate bending at fracture site with minimal callus but no sequestrum (Figure 3). 20^0 angulation and 1.5 cm shortening of left lower limb was found. Full Blood Counts (CBCs), ESR and CRP was done. Haemoglobin 13 gm/dl, WBC within normal range but moderate increased of ESR and CRP.

Figure 2 Follow-up x-ray after 3 months.

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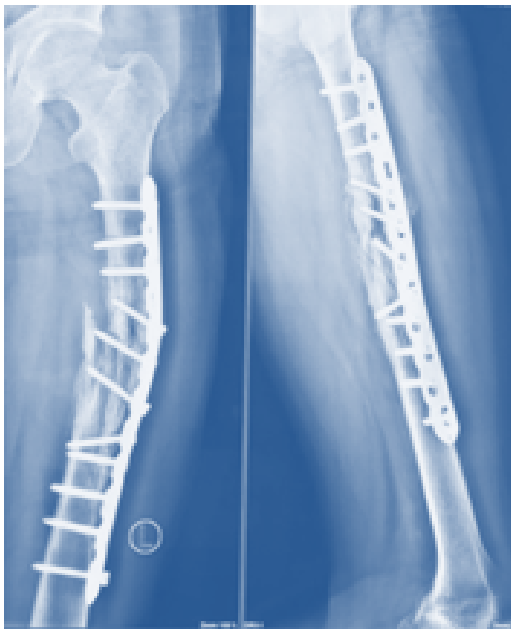

Figure 3 x-ray after re-fracture due to fall.

A working diagnosis was H. Rosen`s type 3 Quiescent infected non-union (Oligotrophic) and Romano stage -1 post-implant infection with re-fracture mid shaft of left femur.

Thorough evaluation of patient was done and after 7 days of re-fracture, modified Ilizarov frame was mounted with plate *in situ* for treatment of the patient (Figure 4, 5).

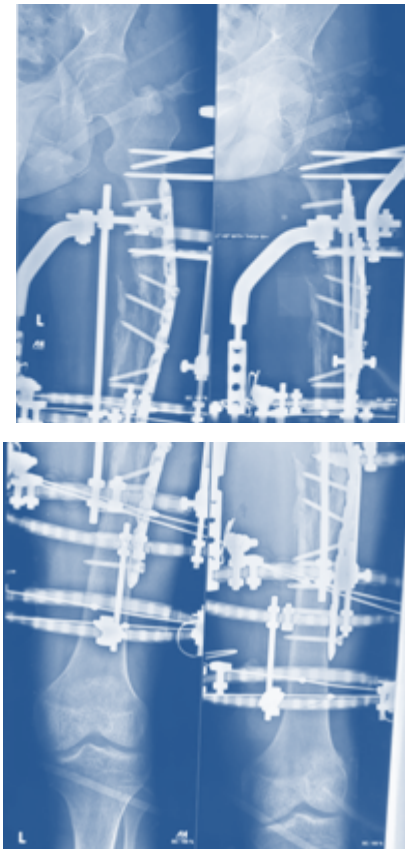

Figure 4 Post-operative x-ray after mount Ilizarov frame kept plate in situ.

Figure 5 Post-operative pictures with Ilizarov frame.

Figure 6 Follow up x-ray after A.M. (1 month later).

Illustrations

During Accordion Maneuver (AM), 1st compression was done at the rate of 0.25 mm 2 times in a day for 7 days following rest for 3 days, then distraction was done at same rate for 7 days due to oligotrophic type of non-union. This is a cycle of Accordion maneuver (AM). When completed 1st cycle rest for 3 days and start 2nd cycle of AM. After 2 cycle of AM, maneuver stopped and kept Ilizarov apparatus in place for new bone consolidation. Final compression was done at the rate of 0.25 mm on every 3rd day for 1 month. Abstain from corticotomy because bone lengthening was not done. Initial limb shortening compensated by shoe raised providing rubberized saddle attached to the left foot.

The patient was practiced to pin site care and discharged from hospital when he became well-trained of the apparatus care and could perform AM himself. A regular follow up once in a week for 1st month then monthly interval till fracture union (Figure 6–8).

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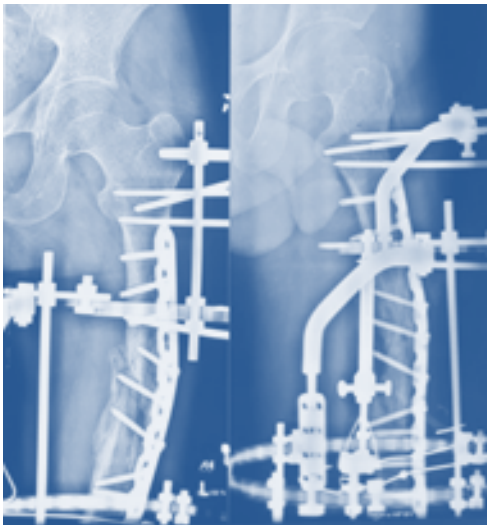

Figure 7 Follow up x-ray after 4 months.

Figure 8 Follow up pictures with Ilizarov frame.

Adamant post-operative protocol was followed with adequate nutrition diet, no smoking, physiotherapy of joints and crutch-assisted walking. Weight-bearing was delayed for 2 weeks. Leg elevated to minimize soft-tissue oedema and delay vascular compromise in large bone gaps that were acutely docked.

After complete healing and consolidation of regenerated bone, substantiated by radiological evidence Ilizarov apparatus was dispelled 6 month later of installation without removal of plate (Figure 9). He was instructed to incur physiotherapy to recuperate range of motion (ROM) at the knee and hip joint.

Figure 9 Post-operative x-ray after removal of Ilizarov frame without removal of plate.

After 4 months of Ilizarov apparatus removal, there were no pin site wounds, ROM at knee was 0-120^0 and normal hip motion. C- reactive protein (CRP) and erythrocyte sedimentation rate (ESR) were done 3 consecutive tests in a week of surgery, the values in normal range and did not increased, also intra-operative bacteriological culture was done to see deep seated infection. There was no evidence of infection. Finally plate was removed. In radiograph there was 15^0 angulation and 1cm shortening of left lower limb. After 6 months radiograph 13^0 angulation. One (1) year and four (4) months radiograph after plate removal, there was bone remodeling and 6^0 angulation with 1 cm shortening of left lower limb found (Figure 10,11).

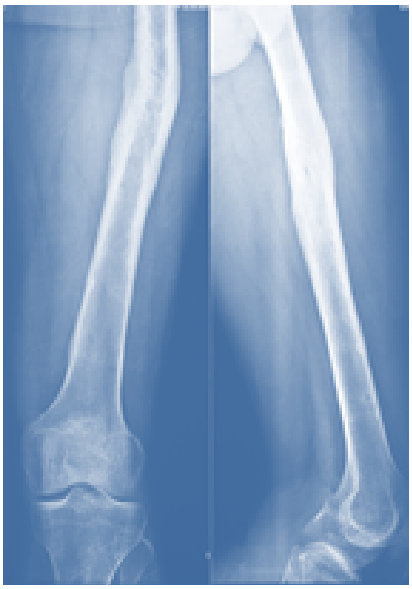

Figure 10 X-ray after removal of plate and consolidation (1 year & 4 months later).

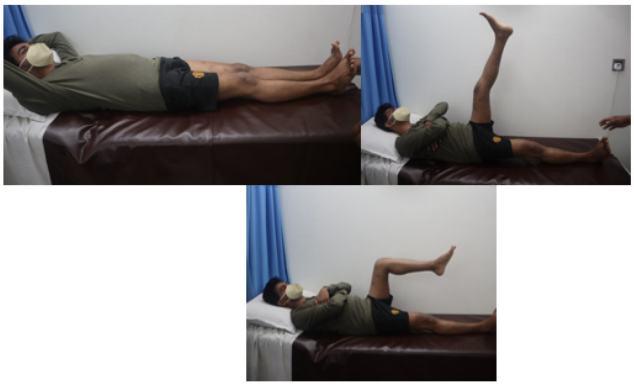

Figure 11 Pictures after full recovery from fracture after one (1) year and four (4) months.

Discussion

H. Rosen`s type 3 Quiescent infected non-union (Oligotrophic) and Romano stage -1 post-implant infection with re-fracture mid shaft of left femur, require no debridement and removal of implant (Plate), therefore can be treated by a single stage procedure.[3]

Femoral shaft fractures (FSFs) are frequently occurring injury, due to high-energy trauma such as Road Traffic Accident (RTA) and also low-energy trauma, like fracture in elderly caused osteoporosis. Femoral shaft fractures are 10 – 21 per 100,000 per year.[4] In the case, Gustilo type-I, comminuted fracture, middle segment shaft of left femur by motor vehicle accident.

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There are many treatment options in fracture shaft of femur but deficiency of standard guidelines. The treatment options are open reduction and internal fixation (ORIF) by plate, intramedullary nail (both open and close methods) or by external fixator. The more diversity of treatment options, reflect more challenges of femoral shaft fracture. Depending upon the surgeons, treatment option may choose on their experience and knowledge and the availability of tools and implants. The more apprehend complication in fracture management is non-union, amalgamated with bone loss, deformity and infection.[9] The gold standard treatment for femoral shaft fracture is intramedullary nailing (IMN) with low post-operative complications – infection rate 1.2 to 5%, non-union 8%, broken nail, reaming trauma and technical difficulties but when plate is used complications are more – wide range of surgical trauma, infection 5%, non-union 19% and broken or bending of plate.[5,10–13]

Closed intra-medullary nail (IMN) fixation provide more benefit of mechanical stability, low bleeding and less surgical damage which reduces risk of infection compared to plate fixation.[10] Plate fixation is suitable in comminuted fracture shaft of femur and minimally invasive plate osteo-synthesis (MIPO) procedure preserve vascularity and soft tissue which facelifted bone healing.

The patient`s fracture was fixed by Broad Dynamic Compression Plate (DCP) with Minimally Invasive Plate Osteo-synthesis (MIPO) procedure three (3) days after trauma. Post-operative radiograph ensured bony fragments and plate was in position, maintaining well alignment with minimal fracture gap between fragments (Figure 1).

We choose MIPO technique for the treatment of the patient due to nature of the fracture (comminuted with large longitudinal and several spiral fracture lines. There were small bony pieces within the bone). Although IMN fixation is the standard for femoral shaft fracture treatment which provide more stability but we avoid nailing because it is very difficult to insert in this type of fracture by close method and need to be open. There was possibility of more spread of fragments during reaming and nail insertion which persist as fracture gap. Also extensive surgical trauma of soft tissue and vascularity was lost. By MIPO we preserve above all things.

Infection and non-union are the morbid complications of femur fractures. Infections are more common in open fracture and also caused by superficial surgical site infection (SSSI). The incidence of infection in closed long bones fractures 1-2% and it is higher in open fracture that is 5% in Gustilo type-I, 10% in Gustilo type-I and 15% in Gustilo type-I. In femoral fractures infection rate is 4-7% in open and 1-2% in closed cases,[14] and surgical site infection (SSI) in femur fractures are 3.6 to 4.8%.[15] The fracture complexity and soft tissue damage are the critical factors, influence the risk of infection. Thorough clinical evaluation, proper investigations and a specific treatment strategy are the troublesome factors to diagnosis infection after long bone fracture surgery. Full/Complete blood counts (CBCs), ESR, CRP with radiological image studies can be give accurate information of underlying infection. Infected tissue culture provides the final and specific diagnosis.[14] Post implant classification is done according to Romano CL.et al. (Table 1)

Stage 1: Wound debridement and surgical toileting. Intravenous antibiotic given as per culture and sensitivity test (C/S). No need implant remove.

Stage 2: Treated with antibiotic with or without implant removal.

Stage 3: Implant remove and successfully treated by external fixator i.e.: Ilizarov.[14] Infection causes delay union or may lead non-union of fracture due to decrease blood flow and tissue damage.

In the patient, during primary management wound care (Gustilo type-I) and temporary immobilization was done. All necessary investigations (CBCs, ESR, CRP and Radiographic image) were within normal range before operation (plate osteo-synthesis). On 14th post-operative day (POD) patient complaints of fever, local pain and swelling. Examined the wound and found local signs of infection (Redness, Oedematous but no discharged). Aspiration was done with wide bore needle, thick pus came out (Table 2). Exploration and surgical toileting was done on same day, wound dressing done daily. Secondary wound closure done after 3 days and intravenous antibiotic continued for 6 weeks. The most common micro-organism in SSI are 20% Staphylococcus aureus, 20% Staphylococcus ssp, 20% Escherichia coli, 40% Enterobacter ssp. and 20% Edwardsiella ssp. The systemic antibiotic therapy and radical surgical debridement established the basis of the treatment of wound infection. The increase of resistant bacteria's, especially the plaque-forming Staphylococcus, has reduced the efficiency of antibiotic therapy. Therefore, surgical debridement has become more important.[15,17]

Table 1 Romano CL.et al. classification of post-implant infections.[3,16]

	Stage 1	Stage 2	Stage 3
Post-operative period	2 to 6 weeks	2 to 9 months	>2 months
Involvement of tissue	Soft tissue	Bone (Non Intra-medullary)	Bone (Intra-medullary)
Removal of implant	Not required	Not required	Required

Table 2 Clinical signs observed in the patients with SSI in femur fracture

Clinical Signs	Patient present
Serous or bloody exudate	No
Edema, hyperemia and local temperature increase	Yes
Purulent, yellowish or greenish exudate	No
Hardened or floating area in place	No
Sensory lowering and vomiting	No
Local pain	Yes

During subsequent follow up wound was healed and infection subsided. According to Romano classification (Table 1) the fracture was in stage - I post-implant infection, as infection was diagnosed within 2 weeks of initial surgery and not spread to bone. The plate provide sufficient stability, That`s why it kept in position.

Re-fractures are another complication by further trauma where implant failure (Broken, Bending, Loosening) in the region of improper callus formation. The incidence of re-fracture is 6.5 to 14.2%.[7] Approximately 5 - 12% non-union occurs in all fracture and it is around 20% for diaphyseal fracture.[18,19] Delayed unions (3%) and non-unions are more common in femur fracture (1.1 - 14%). On average 200 cases of long bone non-union occur per million population, estimating of 150.000 cases in Europe each year.[5,20,21]

The patient came after 4 months of his operation with the complaint of pain in left thigh during walking following trauma by fall. During physical examination we found moderate tenderness without any abnormal mobility at fractured site. There were no signs of infection. Knee and hip joints movement restricted due to pain. In radiograph there was re-fracture of mid shaft of left femur with plate bending at fracture site with minimal callus but no sequestrum (Figure 3) 20^0 angulation and 1.5 cm shortening of left lower limb was found.

Non-unions are invariably occurred with multiple factors being implicated in this incidence. Those are systemic compromise of the host, local condition of the affected area, specific injury characteristics and iatrogenic factors relating to the treatment of the initial injury. [22] Most commonly, inadequate stability, poor blood supply and deep seated hidden infection lead to develop non-union.

Non-union is treating by appropriate mechanical stability, ensure biologically active bone ends, bone grafting, improve blood supply, tissue debridement and antibiotic to eradicate infection. Sometimes increase impression of BMP7, BMP2, TGF-β1and osteoblast proliferation through bone simulation by Direct current (DC), Capacitively coupled electrical field (AC), Pulsed electromagnetic field and Combined magnetic field (Table 3).

Table 3 H. Rosen`s classification of Infected Non-union with pathology and treatment strategy.[3,23]

	Type 1 (Draining)	Type 2 (Active Non-draining)	Type 3 (Quiescent)
Discharging sinus	Exist	Not exist	Not exist > 3 months
Sequestrum	Exist	Usually not exist/small size	Not exist
Extent of debridement	Extensive	Moderate	Nil or minimum when implant has to be removed

Full/Complete Blood Counts (CBCs), ESR and CRP was done. Haemoglobin 13 gm/dl, WBC within normal range but moderate increased of ESR and CRP.

According to H. Rosen`s classification (Table 3) the fracture was type 3 Quiescent infected non-union (Oligotrophic), because there was no discharge from local site for more than 3 months and no evidence of sequestrum. Surgical toileting and least amount debridement was done during exploration.

Final working diagnosis was H. Rosen`s type 3 Quiescent infected non-union (Oligotrophic) and Romano stage-1 post-implant infection with re-fracture mid shaft of left femur.

Bone has intrinsic capability to heal spontaneously following injury. The management of an infected non-union one of the greatest challenge in fracture surgery, not only the surgeon, non-union has a devastating impact on the patient`s function and life quality. Bone infection after fracture treatment and it`s management is a time consuming and challenging procedure for Orthopedics and Trauma Surgeon.[9]

Infected non-union, aseptic non-union, neglected open fracture and neglected displaced comminuted fracture are the difficult fractures of femur.[8]

A variety of approaches have been applied to treat oligotrophic non-union. Conventional methods usually accompanied with large operative trauma, more blood loss, risk of re-infection, higher medical cost and complications at donor site of bone grafting.[18] The suggested treatment for quiescent type of infected non-union is a single stage procedure with minimal or no debridement and if implant is provide sufficient stability it placed *in situ*.[2] The healing rate for femora shaft non-unions is too high (90%).[1]

We were decided for the treatment of the patient, Accordion Maneuver with Ilizarov apparatus, a single stage procedure in femoral shaft non-union and re-fracture with quiescent infection kept plate *in situ*.

In 1950 Professor G. A. Ilizarov, a man of Russian physician introduce a new pioneer concept to treatment of fracture and orthopedics patients, which is memorable as Ilizarov methods. The circular external fixators attached with bone fragments by tensioned wires and rings are connected each other's by rods or telescopic rods. Assembling the ring is the first components that provide stability, protect soft tissue and hold optimum mechanical with biological field for regeneration, remodeling and rehabilitation in the treatment of fracture and orthopedics diseases. [24] Gradual traction on living tissues create stress which stimulate and maintain the regeneration of tissues, called **law of tension-stress**.[25,26]

Ilizarov osteo-synthesis produce tension-stress effect to living tissue causes new tissue formation which is the basic principle of the treatment of many complex injuries and diseases of locomotor system.[27] Living cells become metabolically active by slow and steady traction on it, resulting in increased biosynthetic functions and proliferation of the cells. [25] The processes are depending on the adequate blood supply and the effect of weight-bearing with functional activity. Ilizarov circular external fixators are outline of differing stability of fixation which protects periosteum, bone marrow and blood supply. Fixation stability with preservation of periosseous and intraosseous soft tissues enhance bone formation.

Distraction and transformational osteogenesis are seen in Ilizarov method. Distraction osteogenesis (DO) use for bone regeneration and bone transport (Bone lengthening), whereas transformational osteogenesis use for healing non-union and hardening hypo-regenerate in bone transport. It mechanically stimulates pathologic bony infections to induce osteogenesis and regenerate normal bony continuity. [2]

The new bone formed parallel to the tension vector even when perpendicular to the bone`s mechanical axis. Bone marrow damage averts osteogenesis, occurred by the lateral tension-stress vector.[27]

The quality and quantity of regenerated bone confide by-1. Rigid fractures stability 2. Degree of damage (Soft tissue, bone marrow and Nutrient artery with it`s branches) 3. Distraction rate (Speed) and 4. Distraction rhythm (Frequency),[25,27,28] all of which defend perfectly by Ilizarov apparatus.

Accordion Maneuver (AM)

Accordion Maneuver (AM) is a «Bloodless stimulation″ of tissue regeneration described by Professor G. A. ILIZAROV, which encircled with intermittent compression and distraction like a musical instrument and stimulate tissue neo-genesis and also convert inactive scar tissue into biologically active tissue for regeneration. A modified form of this tool can be apply in hypo-regenerate state, developed in bone transport and docking site after acute docking or after internal bone transport.[2,24,29]

Accordion Maneuver based on the principle of transformational osteogenesis,.[1] which control both healing and shape formation of bone and soft tissues. This bloodless tool helps us to abstain the needs of bone grafting and shaving the bone edges.[8,27]

AM used worldwide to treat a wide variety of musculoskeletal and craniofacial conditions, including angular deformity correction, management of bone defect secondary to infection, trauma or tumor, in non-union site, docking site during bone transport and in hypo-regenerate state of bone healing.[25,27]

Influencing factors of AM

Various host related, local and iatrogenic causes may lead to poor regeneration in AM. These are generalized systemic diseases, infection, immune-suppression, malnutrition, metabolic, reduce vascularity, reduce walk, lack of soft tissue covering, radiation exposer, fixation instability, sub-optimal osteotomy and too fast distraction rate.[2,28] AM fails to stimulate regeneration most likely due to underlying deep seated infection. It is important to judge the risk factors that may causes of poor regeneration in distraction osteogenesis (DO) prior to apply accordion maneuver.

Mechanism of accordion maneuver

Accordion Maneuver works through cyclical use of distraction and compression. Distraction osteogenesis is a biological phenomenon that utilized to induce formation of new bone and soft tissue. Slow and gradual tension on living tissues produce stress to the cell which initiate distraction osteogenesis as a result regeneration of tissue occur. New bone formation in the gap at parallel to the vector of tension. Regeneration of bone continues until tissue is stress incrementally and disruption of the blood vessels in the gap is confined. The increased bones forming activity is imposed to the stimulatory effect of tension on bone forming cells and on blood vessels (facilitate blood flow).[8] There is only one study found, that directly compared the effect of compression versus distraction in regeneration. Hente R. et al. observed the amount of periosteal callus formation was up to 25 times greater on compression side in compare to distraction side, applying a specially designed external fixator.[28,30] This may explain the positive effect of compression during accordion maneuver.

Phases of Accordion Maneuver as well as distraction osteogenesis:[2,3,24.26,29,31]

Latency phase: It is the period (5 to 10 days) delay before distraction. 5 days in child and 10 days in adult. The phase immediately followed to osteotomy and assumes formation and organization of hematoma.

Distraction phase: Bone segments are gradually distracted in various step increased until the desired healing obtained. It creates columnar fibro-vascular tissues that arise from crushed scar tissue on fracture surface. The optimum rate found at 1.0 mm/day by Ilizarov and the optimum rhythm of distraction at 0.25 mm/6 hours.

Compression phase: Temporary cyclic compression and distraction promote bone healing by increasing bone volume that has higher bending rigidity. Compression bring the fragments into close contact and crushes the scar tissue between the fragments.

Consolidation (Neutrofixation) phase: Distraction adjourn and bone fragments are occupied in place until newly formed bone consolidates (About 1 month/CM of callus/ lengthened). Final rhythmical compression leads to consolidation, done at the rate of 0.25 mm on every 3rd day for 1 month.

Removal of the fixator: After complete maturation of newly formed bone by radiological (Bridging of callus across fracture gap) and clinical (Absent of pain during weight bearing) assessment.

Mechanical and cellular event during AM

Cyclical intermittent distraction and compression implicates during AM that initiate regeneration. Osteogenesis occurs by tension-stress, called distraction osteogenesis (DO). DO works through: 1. Mechano-transduction, which change into a cascade of molecular signals and 2. Activate various cellular events; Proliferation, Differentiation and Secretory functions, that ultimately form new bones.

Mechanical events on adaptation of bone structure: Mechano-transduction is the process by which physical forces are converted into biochemical signals that are then integrated into cellular responses. The molecular signaling pathways of mechano-transduction are; Ca++ channel, integrin, wnt/β-catenin, prostaglandin and Nitric Oxide.

Bone repair and regeneration are influenced by interaction between physiological, biochemical and mechano-biological environments. Mechano-transduction in the bone has 4 phases; 1. Mechano-cupling 2. Biochemical coupling 3. Transmission of signals and 4. Effector cell responses. This complex mechanism are responsible for maintaining the dynamic balance between bone formation and bone absorption.

The bone needs "Time off" from mechanical loading. Mechanical loading appear as a potent osteogenic stimulus, but bone cells desensitize promptly to mechanical stimulation. Re-sentization must need before the cells can transduce future mechanical signals effectively. Cyclic or intermittent loading which provide regular "time off" period are more effective than continuous loading in bone regeneration.

Bone consists of, bone forming cell - Osteoblasts (active osteoblast and inactive bone lining cells), bone remodeling cell - Osteoclast and Osteocyte. Osteocytes act as a sensor cell, osteoblasts and osteoclasts are effector cell.

Load to the bone, flow pass to osteocyte processes in their canaliculi, osteocytes can sense the flow of fluid and then produce signaling molecules that generate osteoclast mediated bone resorption and osteoblast mediated bone formation.

Cellular response to mechanical loading depends on: Cell type, Stage of differentiation, Type and magnitude of loading.[29] Fracture healing depending on; 1.Type of fractures 2. Fixation and 3. Loading forces- hydrostatic pressure, tensile strain, shear strain and fluid flow.

Cellular event: Pro-inflammatory cytokines; TNF-α and IL -β, regulate inflammation and participate in bone healing because they are expressed at both very early and late phase of repair process. Cytokines initiate bone healing process and play important role in intra-membranous bone formation and remodeling. Osteogenic protein; BMP2/4, Activator protein genes (AP-1) also osteogenesis.

When mechanical forces apply during distraction, activates time related, local cytokines and growth factors, these are;

- BMP (Bone Morphogenetic Proteins)
- FGF (Fibroblast Growth Factor)
- IGF (Insulin Growth Factor)
- TGF-β (Transforming Growth Factor β)
- PDGF (Platelet Derived Growth Factor)
- VEGF (Vascular Endothelial Growth Factor)
- HIF (Hypoxia Induced Factor) and
- EMPs (extracellular matrix proteins)

Many of those become upgrade during distraction and decline when force stops. DO is a vascular dependent process and new bone formation is associated with robust neo-angiogenesis and neo-vascularity. There is an upward outpouring of many vascular growth factors in the distracted zone including;

- VEGF
- HIF

- Basic FGF and
- Angio-protein

Mobilizations of endothelial progenitor cells play a vital role in neo-angiogenesis and regeneration.[26]

The accordion maneuver also uses to stimulate bone formation in the context of fracture healing.[28] Bone healing is a unique process where regeneration occurred without scar. Both the rate and rhythm of distraction are vital for quantity and quality of regenerated bone. It is well known that the mechanical state plays a leading role in bone regeneration. The bone adapt to the mechanical loads are subjected to in terms of modeling, remodeling and regeneration (Wolff`s law).[28] Experimental studies shows that dynamic compression has greater bone remodeling than static compression, it is due to bone cells requires «time off" from mechanical loading. The bone cells become desensitize promptly to mechanical stimulation and re-sensitization must need before the cell can transduce any prospective mechanical loads into biochemical signals.

Compressive forces may lead to fibro-genesis, osteogenesis and intra-membranous bone formation, while distraction forces may lead to chondro-genesis and endochondral bone formation (Figure 12). Now it is an establish fact that controlled distraction (cyclic alternate compression and distraction) stimulate new bone formation (regeneration) by intra-membranous ossification.[3,29]

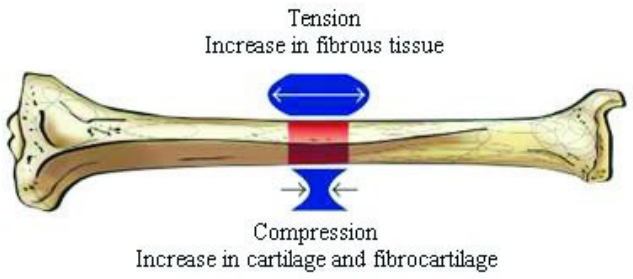

Figure 12 Illustration shows the osteogenic histological outcomes of tension versus compression.[5]

Protocol of accordion maneuver

AM apply when patient become habituated with the Ilizarov apparatus and start mobilization with support. It`s usually 5 to 10 days after mount of ilazarov apparatus (Latency period). Alternate distraction and compression force impose and every step follow by rest, these comprise a cycle of maneuver. How frequent, how long and when these forces should be applied to achieve optimum result, till now remain unanswered question.[28] Distraction or compression, which force apply first depend on non-union type and quality of tissue anticipated between fragment at fracture site. In hypertrophic (Stiff) non-union, distraction followed by compression in each cycle and the steps was reversed in atrophic (Mobile) non-union. In hypo-regenerate condition during bone transport, discontinue distraction and ensure stability, then compression done first, after 2 cycles of AM, check regenerate status by x-ray. After successful maneuver, again start distraction.[2] Previous study on the use of distraction and compression in the treatment of long bone fracture, delayed union and non-union shows the protocol of AM in Table 4 and treatment protocol in delayed or absent callus shows in Table 5.

Baruah and Patowary suggested protocol for the treatment of Postponed Fracture (PF)/Neglected Fracture with Fixation *in situ* (FIS) distraction done 1st at the rate of 0.25 mm, 2 times in a day for 7 days followed by rest for 3–4 days. Then, compression done at the same rate followed by rest. After 2 cycles of AM, final compression was done at the rate of 0.25 mm on every 3rd day for 1 month, [24] and for hypo-regenerate and infected non-union management (B & P) follow Compression for 10 days followed by distraction up to 10 mm - 20 mm at the rate of 0.25 mm 2 times in a day, then rest for 7 – 10 days, after that slow compression again up to 7 mm – 10 mm. After a rest period for 5 – 7 days, distraction is performed for 2nd time. [2,3] Kulkarni suggested protocol for AM in hypertrophic non-union as distraction 0.5 mm/day for 20 days, followed by rest for next 20 days and final compression was done. [2]

Table 4 Previous reports on the use of distraction and compression in treatment of long bone fractures, delayed unions, and non-unions (AM protocol).[28]

Writers	No. of patients	Indication	Effectual outcomes	Procedure of distraction-compression
Kulkarni 2004	N/A	Hypertrophic non-union	N/A	Distraction 0.5 mm/day for 20 days, then stopped for the next 20 days and finally compression
Inan et al. 2005	11	Femoral pseudo-arthrosis	100% (11/11)	Cyclic compression and distraction at the non-union site
Madhusudhan et al. 2008	2	Tibial non-union	100% (2/2)	Compression and distraction (no details)
Laursen et al. 2000	2	Tibial non-union	50% (1/2)	Alternating distraction (1 week) with compression (1 week), until callus found on X-ray
Chand et al. 2010	2	Non-union of long bone fractures	100% (2/2)	Compression and distraction technique (no details)

Table 5 Previously published clinical studies reporting the accordion technique during delayed or absent callus formation of distraction osteogenesis (DO).[28]

Writers	No. of patients	Indication	Effectual outcomes	Procedure of accordion maneuver
Iacobellis et al. 2010	3	Poor regenerate during bone transport	100% (3/3)	Compression followed by distraction for bone transport (no details)
Hatzokos et al. 2011	8	Delayed consolidation	75% (6/8)	Accordion technique (no details).
Kawoosa et al. 2003	1	Delayed consolidation	100% (1/1)	Alternate compression & distraction for re-generate (no details)
El-Mowafi et al. 2005	N = ?	Delayed consolidation	?	Compression & distraction of a moving segment (no details)
El-Sayed et al. 2010	25	Absence of callus formation	76% (19/25)	Distraction & compression technique (no details)
Tsuchiya et al. 1997	N = ?	Poor re-generate during bone transport	?	Compression & distraction for moving segment (no details)

Table Continued..

Writers	No. of patients	Indication	Effectual outcomes	Procedure of accordion maneuver
Vidyadhara and Rao 2007	N = ?	Poor re-generate callus during bone transport	?	Compression & distraction for moving segment (no details).
Simpson and Kenwright 2000	2	Poor callus formation	0% (0/2)	Dynamics change for distraction (no details)
Krishnan et al. 2006	2	Poor regenerate during bone transport	100% (2/2)	Mentioned as distraction discontinued, reversed, and restarted at a reduced rate (0.25 mm/12 h, instead of 0.25 mm/6 h)

? = No. not mentioned

Non-union treatment by accordion maneuver with Ilizarov fixator

In hypertrophic (Stiff) non-union; found thick fibrous, fibrocartilaginous tissue with good blood supply which is biologically active. Distraction 1st started to stimulate neo-osteogenesis followed by compression to consolidate the callus (New bone). In Atrophic (Mobile) and Oligotrophic non-union; loose fibrous tissue is present at the non-union site which is avuscular and biologically inactive, in that case compression done 1st to crush the tissue to invite inflammation and coming neo-vascularization, that is conducive for neo-osteogenesis. Distraction originates columnar fibro-vascular tissues that arise from the crush tissue at fracture site. Repeated distraction stimulates production of osteoblast and helps collagen bundles to consolidate within a bony matrix. [2]

Two (2) types of osteogenesis are seen in Ilizarov technique, distraction osteogenesis (DO) and transformational osteogenesis. Transformational osteogenesis is used for healing non-unions and hardening hypo-regenerate in bone transport. It mechanically excites pathologic bony infections to induce osteogenesis and regenerate normal bony continuity. This is performed by AM as it alternately gives compression and distraction as an accordion. AM is a powerful tool describe in Ilizarov techniques for delayed union, non-union and also in hypo-regenerate treatment during bone transport.

Assessment of Bone deformity, Healing and Time of implant removal

Clinical and radiological assessment was done for notice healing and bone deformity during treatment period. Radiological assessment was done at monthly intervals, starting at the end of 1 month of final compression following two (2) cycles of AM, until union was achieved. Hypo-regenerate state was assessed radiologically at the end of two (2) cycles of AM. It was considered as normo-trophic, if radio-dense new bone appeared, continuity of bone columns was seen. Cross sectional diameter of regenerated bone is equal to width of bone at injury site and central radiolucent band of regenerate tissues was about 4 mm or less. Union was established, if there was bridging callus across fracture site in entire cross section of orthogonal views. [2] Union confirmed clinically by the absence of pain on weight bearing during dynamization. Prior to removal of fixator, dinamization was done by loosening threaded rods, one (1) at a time at weekly intervals.[2,29]

The outcome of Ilizarov treatment was assessed according to ASAMI bone and functional result criteria, also complication (Deformity) was evaluated as per Paley`s classification (Table 6,7).

Table 6 ASAMI Scoring.[17,32,17,18]

Type (Score)	Criteria
Bone results using ASAMI scoring system:	
Excellent	Union, no infection, deformity < 7. LLD < 2.5 cm
Good	Union with any 2 of the following: absence of infection, deformity < 7. LLD < 2.5 cm
Fair	Union with only 1 of the following: absence of infection, deformity < 7. LLD < 2.5 cm
Poor	Nonunion/re-fracture/union with infection, with deformity >7 and LLD >2.5 cm
Functional results using ASAMI scoring system:	
Excellent	Active, no limp, minimum knee stiffness (loss of < 15^0 extension/<15^0 dorsiflexion), No RSD, Insignificant pain.
Good	Active, with 1 or 2 of the following: limp, stiffness, RSD and significant pain.
Fair	Active, with 3 or all of the following: limp, stiffness, RSD, significant pain.
Poor	Inactive (unemployment or inability to perform daily activities because of injury)
Failures	Amputation

LLD, limb length discrepancy; RSD, reflex sympathetic dystrophy

Table 7 Paley Classification of bone deformity.[17,30,32]

Paley type	Deformities
Type I	Intact femur with mobile hip and knee a) Normal ossification proximal femur b) Delayed ossification proximal femur
Type $I-_0$	Ready for surgery, no factors to correct before lengthening
Type $I-_1$	One (1) factor to correct before lengthening
Type $I-_2$	Two (2) factors to correct before lengthening
Type $I-_3$	Three (3) factors to correct before lengthening
Type 2	Type 2: mobile pseudarthrosis with mobile knee a) Femoral head mobile in acetabulum b) Femoral head absent or stiff in acetabulum
Type 3	diaphyseal deficiency of femur a) Knee motion > 45 degrees b) Knee motion < 45 degrees

Factors requiring correction prior to lengthening of femur: NSA < 90^0 +/- delayed ossification proximal femur, CE angle < 20^0 subluxing patella and/or dislocating knee.

Accordion Maneuver (AM) is the "Bloodless Stimulation" of bone healing described by Professor Ilizarov. It comprises of alternate compression and distraction which produce stress in living tissue and also convert biologically inactive scar tissue at non-union site into tissue capable of neo-osteogenesis.[1] The increased bone forming activity as a result of distraction is imposed to the stimulatory effect of tension on blood vessels formation which increases blood follow and regeneration. [26]

Distraction Osteogenesis (DO) is a surgical procedure practice worldwide to give management of a wide variety of musculoskeletal and craniofacial conditions, like- angulation deformity correction and bone defects secondary to infection, trauma or tumor by limb lengthening or segmental bone transport. Although Distraction Osteogenesis (DO) have satisfactory outcomes in most of the cases

but some cases there was absent or delayed callus formation in the fracture gap occurred. [28]

Circular external fixators using Ilizarov`s principles give consistent outcomes in such difficult scenarios, disparate others conventional procedure. It stimulates and initiate neo-osteogenesis without bone grafting and also regenerate soft tissue beside plastic reconstruction surgery. [8]

The aim of treating the case is to acquire healing of the fracture non-union without re-activation of infection. Multiple intervention lead to poor vascularity at the operative site and the stability provided by the earlier implants was not adequate to maintain the alignment till the revascularization process was complete. [29] A less traumatic and minimally invasive single stage procedure would be the best choice of such critical situation. Since there was no evidence of active infection both clinical and in radiograph, we initiated to mount the Ilizarov frame with plate *in situ*. As screws became loosen after re-fracture seen in x-ray, (Figure 3) we were expected the bony fragments could be moved on it during compression and distraction of AM cycles. There was no bone loss, so corticotomy was not done. Thorough evaluation of the patient, after 7 days of re-fracture, modified Ilizarov frame was mounted with plate *in situ* for treatment of the patient.

The Ilizarov apparatus consisted of an arch in the proximal segment at the level of greater trochanter and full frame at lower 1/3rd of femur through two (2) rings. 1.5 mm schanz screws were used in arch and olive wires in distal rings.Figure 4 Accordion maneuver (AM) was started from 7th post-operative day of Ilizarov. We follow R. K. Baruah and S. Patowary protocol of accordion maneuver.

- **The external fixator index** was calculated as external fixator duration (day)/amount of lengthening (cm) or non-union fracture area (cm).*
- **The consolidation index** was calculated as the duration of bone observation at least in 3 cortical layers at the lengthening line (day)/amount of lengthening or non-union fracture area (cm).*
- **The union at the fracture line** was defined as the union and callus tissue formation in 3 out of 4 cortex at the fracture ends.

In the subsequent follow we found –

After clinical and radiological assessment confirmed bony union, after that start dinamization. After complete healing and consolidation of regenerated bone, substantiated by radiological evidence Ilizarov apparatus was dispelled 6 month later of installation without removal of plate (Figure 7). He was instructed to incur physiotherapy to recuperate range of motion (ROM) at the knee and hip joint.

After 4 months of Ilizarov apparatus removal, there were no pin site wounds, ROM at knee was 0-120^0 and normal hip motion. C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) were done 3 consecutive tests in a week of surgery, the values in normal range and did not increased, also intra-operative bacteriological culture was done to see deep seated infection. There was no evidence of infection. Finally plate was removed (Figure 8). In radiograph there was 15^0 angulation and one (1) cm shortening of left lower limb. After 6 months radiograph 13^0 angulation. One (1) year and four (4) months after plate removal by bone remodeling there is 6^0 angulation in radiograph and one (1) cm shortening of left lower limb (Figure 8).

Results

After complete union and consolidation of regenerated bone, substantiated by radiological evidence Ilizarov apparatus was dispelled 6 month later of installation without removal of plate and 4 months of that finally plate was removed.

Table 8 Bone healing status of the patient.[17]

Criteria	Patient present
External fixation time (months)	6 months
Consolidation time (months)	10 months
Union time (months)	12 months
External fixator index	15 day/cm *
Consolidation index	30 day/ cm *

* Instead of lengthening we calculate by non-union fracture area (12 cm)

Table 9 Bone defect status of the patient.[17]

Criteria	Patient present
Femoral defect at first (cm)	Nil
Bone resection (cm)	Nil
Angulation after ilizarov fixation	15^0
Amount of elongation (cm)	Nil
Shortening at the end (cm)	1 cm
Follow up (months)	18 months

Table 10 ASAMI criteria, bone and functional status, also complication classification of the patient.[17,30,17,18]

Criteria	Patient present
ASAMI bone	Excellent *
ASAMIfunction	Excellent **
Knee flexion	135^0 (Full flexion)
Knee extension loss	0^0 (No loss)
Paley 1	Intact femur with normal ROM of knee and hip joints
Paley 2	None
Paley 3	None

+ Bone union, No infection, Angulation -6^0, LLD – 1cm

** Active, No limp, Full ROM of knee (flexion -135^0 and extension -0^0), No RSD, No pain

After one (1) year and four (4) months of mounted Ilizarov frame, now the patient are full free movement of knee and hip. He had no question during walking even running.

Conclusion

Few authors reported, Accordion Maneuver (AM) technique with Ilizarov apply over intramedullary nail (IMN) *in situ* for aseptic non-union of femur. In this study, we discussed the role of this tool while treating femoral shaft non-union and re-fracture with quiescent infection place dynamic compression plate (DCP) *in situ*.

After a thoroughgoing literature review, we are not bringing out any similar case, which make our case as an empirical innovation. We assess the results of Accordion Maneuver with modified Ilizarov osteo-synthesis in the case and highlight their relevance in present orthopedics practice where multiple newer choices have been become famous.

Consent

We are certifying that; We have taken all appropriate patient`s consent forms. In the forms, the patient given his consent for use of his images (both photographic and radiological) and clinical information to be published both in electronic and print form. The

patient understand that his name and initials will not be published and due efforts will be made to hide his identity.

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Conflicts of interest

There is no conflict of interest.

Authors contributions

C. F. Rob was leading and major contribution in writing and editing the manuscript, J. Hossain was main contribution in writing and editing the manuscript, literature reviews, analyzed and interpreted data regarding the case, Md. Gulam Mustofa data collection and literature reviews.

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