



Spina bifida neuropathic foot osteomyelitis : results of using ilizarov technique in 46 cases

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Neuropathic foot osteomyelitis is a challenging condition for both surgeon and patient, we retrospectively reviewed patients with spina bifida who had chronic foot osteomyelitis presented to our centre from 1995 to 2016. The study yielded 46 patients with chronically infected and deformed feet with a mean age of 31.26 years (range : 18-70 years ; SD : 9.61) and Mean follow-up of 5.76 years (range 1-21years ; SD : 3.9). Our technique included a staged debridement IV chemotherapy and ilizarov fixation followed by arthrodesis in 44 cases and calcaneisation in two cases. American Orthopedic Foot and Ankle Association (AOFAS) score was used to assess the results with a Pre-operative mean of ($72.73 \pm SD10.11$) and a value of ($82.32 \pm SD 7.5$) at final follow-up respectively, P-value was 0.29. Laboratory, x-ray, CT scans and foot prints were used to assess pre and postoperative results. Overall 76% had good outcome with a plantigrade stable united and infection free feet. Those with recurrences were treated with the same technique and achieved the final goal of treatment. Using ilizarov technique in the treatment of neuropathic spina bifida foot osteomyelitis presents a reliable and more suitable option for those patients with infected deformed and stiff feet.

Keywords : Spina bifida ; chronic osteomyelitis ; foot ulcers, ; foot deformity ; Ilizarov technique.

INTRODUCTION

Spina bifida is one of the most common birth defects (23) resulting from a defect in the posterior elements due to myelodysplasia, this dysplasia of the spinal cord and nerve roots leads to bowel, bladder, motor, and sensory paralysis below the level of the lesion as well as congenital and acquired orthopaedic deformities (28) congenital deformities includes clubfoot and vertical talus while acquired developmental deformities are caused by muscle imbalance, paralysis, and decreased sensation in the lower extremities like cavovarus, clubfoot, vertical talus, calcaneovalgus and equinus (26,8). Peripheral neuropathy together with muscle imbalance and disturbed biomechanics with oblique axis of the foot disrupt the normal structure of the plantar skin leading to fissuring and sloughing and makes it prone to pressure sores which are recurrent and

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non-healing (11), progression of osteomyelitis may lead in severe cases to amputation (22) the resulting neurogenic deformities usually requires some joints fusion due to muscle imbalance (11,22,26). when infectious agents penetrates the tissues, there is no adequate blood flow to the interstitial tissues and wound area, which contributes to progression of infection, the resulting foot osteomyelitis is potentially devastating condition with diagnostic and treatment challenges, there are many surgical techniques used for management (4,6,7,9,12,13,14,15, 19,20,21,25). Sequestrectomy with debridement of all non-viable tissues remains a main operation that treat osteomyelitis in most cases, it is worth noting that a limited Sequestrectomy causes a high rate of recurrence (10). Optimization of blood supply, soft tissues, limb biomechanics, and the physiologic state of the patient combined with culture specific anti- microbial therapy must be considered as a gold standard approach for treatment of such conditions (2,7,10,14,17,25,29). This study report the outcome of use of debridement, IV etiotropic antibiotics, ilizarov frame fixation and arthrodesis in treating neuropathic spina bifida foot osteomyelitis.

MATERIALS AND METHODS

This is a retrospective review of all patients with Spina bifida who had foot chronic osteomyelitis and deformities presented to our center between 1995 and 2016, there were 46 patients (23 males, 23 females), diagnostic and treatment techniques were reviewed, The mean age of patients was 31.26 years (range : 18-70 years ; SD : 9.61). Mean follow-up time was 5.76 years (range 1-21 years ; SD : 3.9). 26 patients had right, 17 left while 3 had bilateral feet infection. Statistical analysis was calculated using Excel package 2010. Patient's parameters and values were calculated as mean \pm standard deviation, a p-value less than 0.05 was considered significant. The study was performed with approval of the ethics committee (Institutional Review Board), Informed consent was obtained from all participants in the study, out of the 46 patients there were 18 patients with grade C and 28 with grade D, according to the American Spinal Injury Association (ASIA). The preoperative and postoperative American Ortho-

Table I.— Patients duration of deformity and osteomyelitis

| Duration | <5 years | 5-10 years | >10 years |
|---------------|-------------|-------------|-------------|
| Osteomyelitis | 15 patients | 18 patients | 13 patients |
| Deformity | 2 patients | 17 patients | 27 patients |

Table II. — Size and location of ulcer

| Location | | | | | |
|---------------------|-------|------|-----------|-------|------|
| Forefoot – Mid foot | | | Hind foot | | |
| <3cm | 3-8cm | >8cm | <3cm | 3-8cm | >8cm |
| 11 | 9 | 2 | 6 | 14 | 4 |

pedic foot and ankle association (AOFAS) score was assessed in all patients. When first seen all patients had established chronic osteomyelitis and had previous failed operations and had multiple wound debridement and antibiotics, 28 patients received multiple operations mainly debridement and oral or IV antibiotics, 13 received only antibiotics and 5 had no treatment. With regard to foot deformity 20 patients had equino-varus, 9 calcaneus, 7 cavus, 7 varus and 3 valgus feet. duration of osteomyelitis and deformity can be seen in (Table I). with regard to the location of osteomyelitis 22 were in the 4th and 5th metatarsals, 18 in the calcaneum, 4 in the calcaneum and talus together and 2 in the talus. We used Cierny-Mader staging system for osteomyelitis (5) to assess the extent of osteomyelitis. foot ulcers and their size and location were assessed clinically (Table II), a swab for culture and sensitivity was taken preoperatively, another intra-operative swab is taken for confirmation, early results usually appear after 3 days and final results after 5 days, empirical antibiotic covering both gram positive and negative is administered intra-operatively, cefazolin 2g/8 hours and ciprofloxacin 200mg/12 hours are given together. When the results of the first and second swab appear, the patient will be given the culture-sensitive antibiotic for more than two weeks.

Laboratory results of our patients before treatment were typical for a chronic osteomyelitis, there complete blood count showed an increase in leukocytes, acceleration of ESR, and low hemoglobin. Given the presence of concomitant pelvic organs problems especially the bladder a general analysis of urine often revealed proteinuria and leucocyturia. Biochemical blood tests before treatment were characterized by an increase in the

level of C-reactive protein and a decrease in the total protein content. CT scans and plain x-rays as well as footprints were used to assess pre and postoperative status of the affected foot.

Treatment technique

Our treatment plan consisted of two stages, the first stage debridement, partial deformity correction and ilizarov fixation with Intravenous antibiotics and the second stage consisted mainly of completing deformity correction which can be acute or gradual depending on the degree of the deformity and flexibility of the foot followed by Panarthrodesis to stabilize the foot and bone transport when necessary for calcaneisation. Intra-operative swabs are taken from fluid, superficial and deep tissues and bone, we review x-rays, CT scans and when there is sinus we inject a brilliant green dye to determine the extent of the infection, skin and soft tissue incisions are made layer by layer to access bone segments, removal of infected tissues is done until healthy bleeding

margins could be noticed on the osseous bed, when sequestrectomy is performed, the wound is washed thoroughly with antiseptic solution and with ultrasonic wash system, a drain is inserted inside the wound and with skin undermining a primary closure of the wound is done. The foot is then brought to a corrected position that is maximally advantageous for wound healing. In the postoperative period the patient receives a course of etiotropic antimicrobial therapy for two weeks, at this stage the fixation of the foot after the operation lasts for 25-30 days. The extremity is then stabilized by an Ilizarov external fixator (Fig. 1) The frame is constructed by two distal tibial rings connected by rods and a foot half ring connected to two k-wires that passes through 1st and 2nd metatarsal and a nother on that passas through 3rd, 4th and 5th metatarsals, then they are attached to the leg frame by hinged rods , this construct is made for forefoot infections (Fig. 2.a). In case of mid or hind foot infections an additional calcanean half ring connected to the calcaneum by two k-wires is then attached to a forefoot half ring

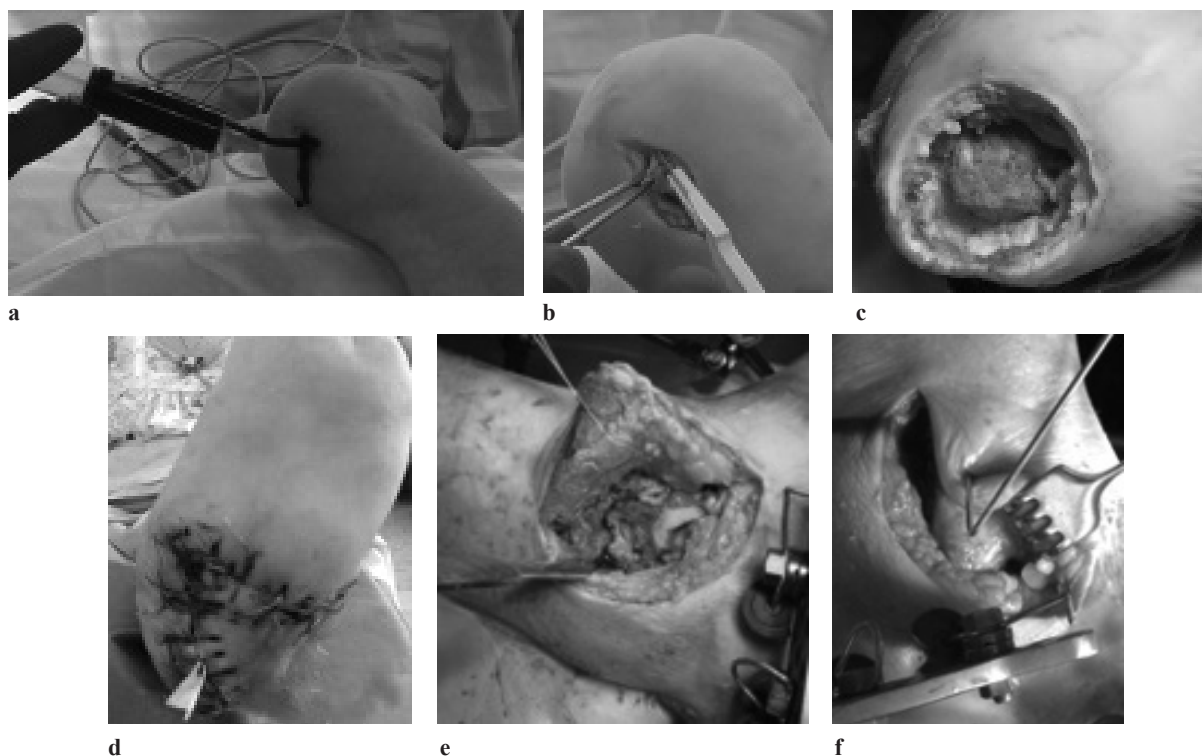


Fig. 1. — Surgical techniques (a) Green brilliance injection (b) sinus excision (c) debridement down to bone (d) Primary skin closure (e) incision for pan-arthrodesis (f) skin closure and wires insertion to complete frame assembly.

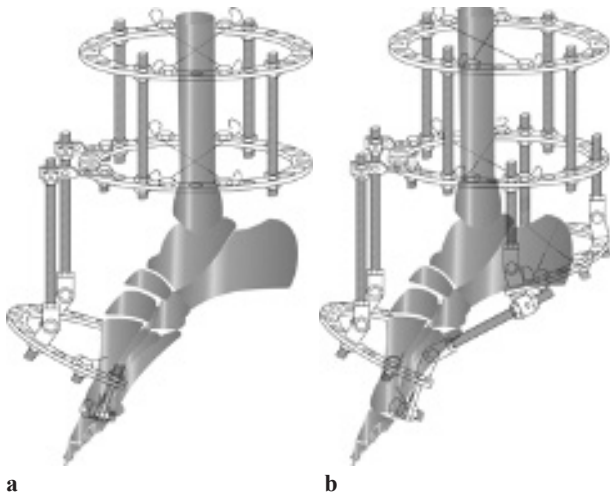


Fig. 2. — Frame assembly in treatment of forefoot infections
(a) Frame for mid and hind foot infections (b).

and connected by rods to the leg frame (Fig. 2.b). Bone defects are filled by soft tissue granulation, more correction is carried out during the second phase of panarthrodesis which begins by performing corrective osteotomies to foot deformities after infection has stopped, distal tibial bone transport to fill calcanean defect (Calcanisation) was carried out in two patients. None of our patients needed skin or soft tissue coverage for their wounds.

Postoperative course

Patients are advised during the first month to avoid loading the foot to allow time for granulation tissue to fill the wound, at the beginning of the

second month the patient is recommended gradual increase of load on the foot. Such regime is maintained until ilizarov frame removal. In our study no patient needed pain medication as all are neuro-pathic. wound dressing is done with gauze soaked in antiseptic solution daily for 5 days, then every other day until end of the second week, then the dressing is carried out once weekly until frame removal. In cases of infection around the wires, dressing was done with rifampicin solution, and gauze is changed daily with local injection of 6mg of Lincomycin once for three days.

After frame removal patients are recommended to wear ankle foot orthosis (AFO) or custom made orthotics for 1 month with gradual increase of load with crutches. Also patients are recommended to do massage sessions to enhance blood supply to the limb, and physiotherapy exercises to develop a new gait pattern after a long standing period of infection and deformity.

RESULTS

The results of Cierny-Mader staging system for osteomyelitis (5) was mainly superficial and then localized and medullary and lastly diffuse in that order (Table III) although most isolated microbe was *Staphylococcus aureus* pre and intra-operatively, the sample also included other recognizable species like *Pseudomonas aeruginosa*, and rare species (Table III) which reflects the diversity of the causative organism that causes chronic osteomyelitis in neuro-pathic feet ulcers. There was no much difference

Table III. — Cierny-Mader staging system for osteomyelitis

| | Stage | Name | Characteristics | Number of patients |
|-------------------|-------|--------------------------------------|--|--------------------|
| Anatomic type | 1 | Medullary | Infection restricted to the bone | 10 |
| | 2 | Superficial | Infection restricted to outer cortex | 18 |
| | 3 | Localized | Well demarcated, full- thickness lesion without instability | 12 |
| | 4 | Diffuse | Infection spread to entire bone circumference with instability | 6 |
| Physiologic class | A | Normal host | No comorbidities | — |
| | B | Bs | Systemic compromise | — |
| | | Bl | Local compromise | 46 |
| | | Bls | Systemic and local compromise | — |
| | C | Prohibitive/poor Clinical conditions | Treatment has a higher risk than Osteomyelitis itself | — |

Table IV. — Pre- and intra-operative microbes isolated

| Preoperative isolated microbes | Postoperative isolated microbes |
|-------------------------------------|--|
| Pseudomonas aeruginosa -11 Patients | Pseudomonas aeruginosa - 10 Patients |
| Staphylococcus aureus - 17 Patients | Staphylococcus aureus - 26 Patients |
| Enterococcus faecium - 6 Patients | Enterococcus faecium - 5 Patients |
| Myroid essp - 1 Patient | Alcaligen essp. - 1 Patient |
| Acinetobacter - 4 Patients | Myroid essp - 1 Patient |
| Proteus - 1 Patient | Acinetobacter - 2 Patients |
| Streptococcus - 5 Patients | Proteus - 1 Patient |
| Pasteurella multocida - 2 Patients | Streptococcus - 5 Patients |
| Serratia spp - 2 Patients | Pasteurella multocida - 1 Patient |
| Klebsiellasp - 2 Patients | Serratia spp - 2 Patients |
| E.coli - 2 Patients | Klebsiella spp - 2 Patients |
| Corynebacteria - 5 patients | Prevotella bivia - 1 Patient |
| Morganella morg -1 patient | Raoultella ornithinolytica - 1 Patient |
| Providencia rettgeri - 1 patient | Vibrio vulnificus - 2 Patients |
| Enterobacter -2 patients | E.coli - 4 Patients |
| Burkholderia cepacia -1 patient | Corynebacteria - 4 patients |
| Klesiella pneumoniae - 5 patients | Morganella morg - 1 patient |

between pre and intra-operative swabs (Table IV). The reconstructive technique involves multiple osteotomies of the foot bones to correct deformities and panarthrodesis to a plantigrade position. the mean total time in external fixator from application to removal was 145.15 days (range 118-161) in arthrodesis group and 258.8 days (range 185-293) for the deformity correction and arthrodesis patients including 2 patients with calcaneisation and five patients which needed additional time in frame to redirect the talus downward when there is > 50% calcaneotomy.

The Pre-operative and postoperative American Orthopedic Foot and Ankle Association (AOFAS) Score was (72.73 ± 10.11) and (82.32 ± 7.5) at final follow-up, P-value at 0.29 which is not significant and student's T test of 6.68, since all patients are neuropathic with senseless feet the pain score will not change very much pre and postoperatively and together with the stiff feet will contribute to the small change in the functional score and an insignificant P-value, but patients reported satisfactory results with correction of deformity

and most importantly arrest of osteomyelitis (Fig. 3). This was achieved by pan-arthrodesis in 44 patients and with calcaneisation in 2 patients. We had 4 cases of recurrence of deformity and 3 cases of infection relapse after one year of treatment, we also had 3 cases of recurrence of deformity and 1 case of infection relapse after 5 years of treatment, so totally we had 7 recurrent deformities and 4 infections. In cases of relapse of deformity, another pan-arthrodesis was carried out with fixation by Ilizarov frame and an extension of frame time to 5-6 months. In cases of recurrence of the osteomyelitis process, the same procedure of debridement was carried out as described above plus extended period of IV antibiotics of 4 weeks, all recurrent cases were treated with no more recurrence at final assessment.

DISCUSSION

Neuropathic ulcers specifically those associated with Spina bifida, Cerebral Palsy, spinal cord lesions, peripheral nerve injuries and leprosy heals slowly and recalcitrant (2,15) an usually complicated by

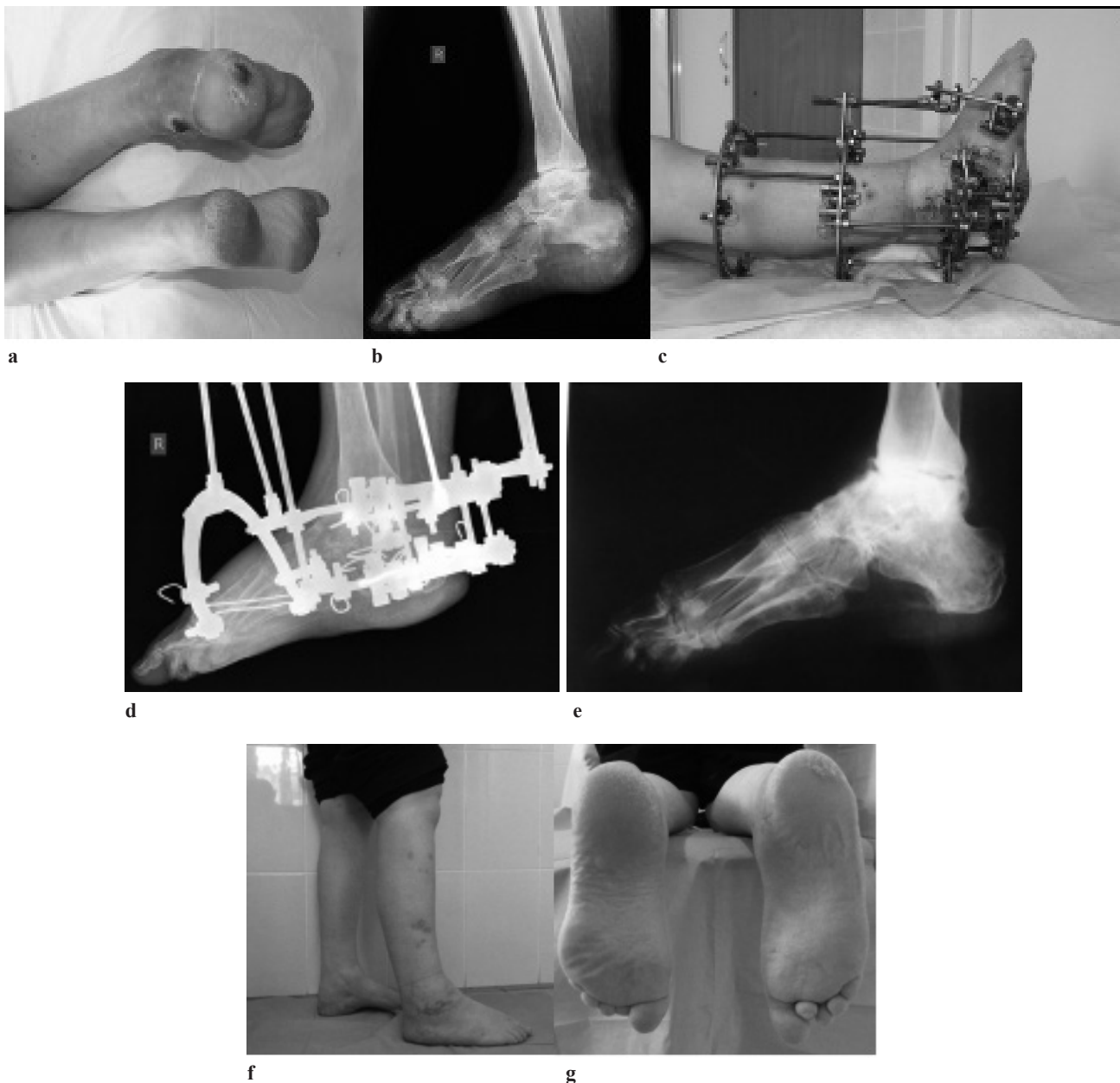


Fig. 3. — 59 years old woman (a) Preoperative right foot ulcers (b) lateral x-ray showing calcaneal osteomyelitis (c) Ilizarov frame during treatment (d) x-ray showing frame assembly for Panarthrodesis (e) three years postoperative x-rays showing bony fusion (f) Patient standing with palmtigrade foot (g) three years with no signs of ulcers or infection.

osteomyelitis that develops secondary to spread of the infection locally into the underlying bone due to decreased blood flow, unstable and deformed joints (2,16,18,25) these effects makes the limb vulnerable to amputation (25). Treatment of osteomyelitis remains a big challenge and requires meticulous debridement,

bone sampling for microbial and histo-pathological examination to allow targeted antimicrobial therapy (10,16). The structural deformity of the foot causes increased pressures on bony prominences on the plantar surface and on an insensible foot which results in ulceration (18,20). In long standing cases

surgical intervention is necessary to obtain a viable vascularized environment and eliminate dead bone (2,4,10,12,14,15,16,17,25). The management of chronic osteomyelitis of an infected foot is complicated by the presence of necrotic bone in a scarred soft tissue envelope and a poor blood supply (5). debridement of all devitalized tissue and foreign material remains the most critical factor in successful management of chronic osteomyelitis (10,27) Henke PK et al (10) did a study including 51,875 patients with chronic osteomyelitis, they concluded that aggressive surgical debridement is vital for limb salvage, In contrast antibiotic therapy alone worsened the condition. While Gitelis S et al. (7), reported using a biodegradable antibiotic-impregnated implant for treatment of chronic osteomyelitis, we managed dead space by packing, drainage and change of dressings until wound is filled by granulation. Lang-Stevenson AI et al. (15) reported treatment of 26 neuropathic ulcers where they emphasized the role of non-weight bearing to off pressure on the healing ulcer. Oganessian OV et al. (19) obtained 71% good results using hinged distraction external fixator to correct 65 (83 feet) equinovarus deformity secondary to ischemic and neuropathic changes due to different causes in all cases a plantigrade foot was obtained. Pieter D'H OOGHE et al. (21) reported successfully treating both skin and bone defects using ilizarov technique with additional rods to approximate the skin edges and cover the defect. Interestingly J correlet al. (12) reported using only ilizarov frame and single dose of intra-operative antibiotic without necrectomy to treat long standing chronic osteomyelitis in 200 cases, 11 of these were in the foot, he concluded that this should be the treatment of choice in severely involved cases with osteomyelitis and chronic ulceration, especially in patients with neuropathic osteomyelitis, e.g. spina bifida, Which support our technique.

Many other authors report high union rate using similar ilizarov and other hybrid external fixators technique to achieve union in different complex foot infection and deformities (4,9,13,19). Even in revision cases, the success and suitability of external fixators use was reported by Easley ME et al. (6) where in revision of tibio-talar arthrodesis the overall union rate for ring external fixation, including revision

and re-revision tibiotalar arthrodesis, was 84.6% (22 of 26) such results were achieved also by Hawkins BJ et al (9) with 21 cases of complex distal tibial pathology or failed ankle arthrodesis where 16 patients (80%) achieved good results with solid ankle arthrodesis. Kołodziej Ł et al. (14) emphasized the role of staging treatment of neurogenic cavovarus feet, which is in agreement with our treatment methods. Calcanean osteomyelitis represent a very challenging situation to treat with partial, total calcanectomy and trans-tibial amputation (1,3) offered as options for treatment. Akkurt MO et al. (1) with partial calcanectomy and ilizarov frame fixation were able to eradicate infection in 18 out of 23 patients (78%) of neuropathic feet. In our sample we had two patients with total calcanectomy, after we treated infection, we managed this large defect by lengthening the distal tibia and redirect it distally and backward down to the plantar surface (calcanisation).

Although many authors (2,4,13,14,16,17) recommend 6 weeks of antibiotics, our patients responded with a 2 to 3 weeks of IV antibiotics, we believe that this result is mainly due to meticulous debridement and etiotropic antibiotics and the use of ilizarov external fixator. Minor complications like pin tract infection were not observed due to our rigorous dressing regime, major complications included recurrence of 7 deformities and 4 infections. All of which were treated successfully without recurrence at the final follow-up.

Limitation of our study is that we could not follow all patients for long time with a mean of follow-up of 5.76 years. Since most of our patients reside in distant areas we advice them to report if there is any problem or recurrence, this proved to be effective in reporting of the recurrent cases. After all, our enrolled patients also functioning stable and without infection feet with full weight-bearing on final assessment. One of the main advantages of external fixator use is the possibility to simultaneously correct deformities and achieve arthrodesis with decreased incidence of recurrence of infection associated with internal devices in such cases.

CONCLUSION

Overall we had 76% good outcome with a plantigrade stable united and without infection or deformity recurrence. Those with recurrence were treated without complications and achieved the final goal of treatment. We believe that staged approach to neuropathic foot osteomyelitis with debridement, deformity correction and IV antibiotics followed by arthrodesis using ilizarov technique present an effective, reliable and more suitable option for neuropathic foot osteomyelitis

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