

Success Story of Ilizarov in Chronic Non-union of Tibia with 18 Years of Morbidity: A Case Report

Ranjit Kumar Baruah¹, S Shyam Sunder¹

Learning Point of the Article:

Role of Ilizarov fixator in salvaging a limb otherwise requiring amputation.

Abstract

Introduction: When the fracture fails to heal for more than 12 months, it is called chronic non-union. Surgical intervention becomes mandatory to get this type of fracture healed. We are reporting our results in a case of congenital pseudoarthrosis of the tibia (CPT) having chronic non-union of 18 years duration with superadded infection following attempted surgery, using Ilizarov technology. We shall highlight the problems found during the course of treatment and different strategies to tackle them.

Case Presentation: An 18-year-old female, born with CPT of the right tibia with neurofibromatosis, presented as Type 1 infected non-union following three unsuccessful surgeries. She had neurofibromatic nodules all over the body including the face. The leg had multiple active draining sinuses, with evidence of healed sinuses and operating scars, adherent to underlying bone. She had 20 cm of limb shortening. Prolonged morbidity made her socially isolated. Being poor, they could not manage her medical expenses. They started believing that amputation would be a viable option and with that feeling in mind, they approached us. After few sittings of counseling, they somehow agreed to give it a try. After preparing the patient, a radical excision of the infected and dead tissue was done. A 3 ring Ilizarov assembly was mounted as the distal fragment was very small. A corticotomy was done to fill up 10 cm of gap following excision and subsequent limb lengthening. She developed hyporegenerate, pin-tract infections (PTIs) during the course of treatment which were tackled accordingly. She also developed psychological problems due to nature of treatment that was taken care of too. The fracture united and she could start walking unaided. There was no recurrence of infection. The ring was kept in situ for about 700 days.

Conclusion: Chronic non-union associated with neurofibromatosis and infection is a very challenging situation. Radical debridement is the key to prevent recurrence of both. Ilizarov system plays the most significant role in forming regenerate by distraction. Hyporegenerate and PTIs could be tackled meticulously to prevent complications and attain the desired result. The patient could achieve painless unaided walking after the treatment. Probably instilling self-confidence in the patient was one of the most important aspects to sustain such an arduous journey.

Keywords: Chronic non-union, neurofibromatosis, infected non-union, Ilizarov.

Introduction

A chronic non-union is defined as a fracture that has failed to heal for more than 12 months, using clinical and radiographic criteria, at which time the non-union diagnosis is not in doubt [1]. Patients diagnosed with non-union are thought to have a very low probability of healing without intervention, and this value

approaches zero for chronic non-unions [2]. There is only one report of chronic non-union of more than 10 years of duration so far [3]. We are reporting our results in a case having chronic non-union of 18 years duration using Ilizarov technology. We will highlight the problems, obstacles, and complication and the different strategies to tackle them.

Access this article online

Website:
www.jocr.co.in

DOI:
10.13107/jocr.2021.v11.i07.2294

Author's Photo Gallery



Dr. Ranjit Kumar Baruah



Dr. S Shyam Sunder

¹Department of Orthopaedics, Assam Medical College Hospital, Dibrugarh, Assam, India.

Address of Correspondence:

Dr. Ranjit Kumar Baruah,
K-Lane, West Milan Nagar, Dibrugarh- 786003, Assam, India.
E-mail: rkb7887@gmail.com

Submitted: 24/02/2021; Review: 05/06/2021; Accepted: June 2021; Published: July 2021

DOI:10.13107/jocr.2021.v11.i07.2294

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License <https://creativecommons.org/licenses/by-nc-sa/4.0/>, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms





Figure 1: Clinical picture of the patient showing neurofibromatic nodules.



Figure 2: Clinical picture showing affected limb with infected sinuses and shortening.

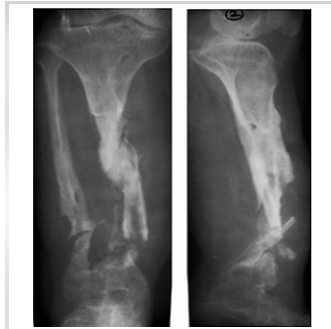


Figure 3: Pre-operative X-ray showing infected non-union right tibia.



Figure 4: X-ray immediately after thorough debridement.

Case Presentation

Demography

An 18-year-old female was born with congenital pseudoarthrosis (CPT) of the right tibia with neurofibromatosis. She underwent three unsuccessful surgeries before presenting to us as Type 1 infected non-union [4] of tibia with shortening at the age of 18 years. She had neurofibromatic nodules all over the body including the face (Fig. 1). The leg had multiple active draining sinuses, with evidence of healed sinuses and operating scars, adherent to the underlying bone (Fig. 2) [5]. There was minimally painful abnormal mobility at the fracture site. The X-ray showed multiple diaphyseal sequestra with cavities, apparent fusion of the ankle joint and atrophic non-union at the distal third of tibia and fibula (Fig. 3) [5].

Prolonged morbidity made her socially isolated. She belonged to a poor family. Meeting her medical expenses became a great burden to the family. The guardians were true to their thinking that no modern science could salvage her limb. They started believing that amputation would be a viable option and with that feeling in mind, they approached us. The clinical setting was also conducive, in general for such an action. We knew that still a modern orthopedic technology called Ilizarov could settle down the morbidities in her favor. However, it was difficult to make them believe that a miracle might still be happening. After few sittings of counseling, they somehow agreed to give it a try.

Treatment given

After improving the general condition of the patient, extensive debridement was done. All dead tissues including bone, soft tissues, and sinuses were excised (Fig. 4). An all-wire Ilizarov frame was mounted with two rings on the proximal fragment and one ring in the small distal fragment. Bone was so osteoporotic that wires could be put by pushing with hand only. The fibula was left untouched. The wound was kept open under massive dressing to heal by secondary intention. The immediate post-operative period was uneventful. A corticotomy was done in the proximal metaphyseal area when the wound appeared healthy after about 2 weeks. Distraction was started after a latency of 7 days and an X-ray on the 10th day of corticotomy showed normal distraction (Fig. 5) [5]. Subsequent X-rays at 1 month interval showed hyporegenerate (Fig. 6). Since the bone was grossly osteoporotic, instability was thought to be the cause of hyporegenerate. One more wire was added in each fragment and the apparatus was further reinforced with two long extension plates. Weight-bearing was encouraged with shoe raise. Two cycles of Accordion maneuver (AM) as per Baruah and Patowary protocol [6] were done at the hyporegenerate site. The wires were tightened in each follow-up. The distraction rate was slowed down to half a millimeter a day in the initial period when bone failed to appear in the beginning.

Once the docking site was reached further distraction was continued to get few millimeters of invagination into distal fragment to improve stability at the docking site. Slowly the

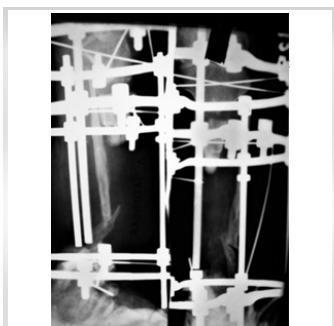


Figure 5: X-ray 10 days after corticotomy.



Figure 6: X-ray showing hyporegenerate at distraction site.



Figure 7: Final X-ray after ring removal.



Figure 8: Final clinical image showing weight-bearing on affected limb.

bone started showing in the regenerate in follow-up X-rays. After docking was achieved, further distraction was continued to gain the length of the limb by external bone transport. Apparatus was kept twice the period of distraction to consolidate the regenerate. It was removed when clinically there was no pain, abnormal mobility, and tenderness, and the X-ray showed three cortices (Fig. 7) [5]. A walking patella tendon bearing cast was applied for 6 weeks to further consolidate the regenerate. She was allowed unaided full weight-bearing afterward (Fig. 8).

Results

The total period of non-union was 18 years as she had CPT since birth and duration of infected non-union was 5 years that she developed after the last operation. The leg was 20 cm shorter than the normal side at presentation. At the final result, she had 2 cm of persistent shortening of the limb. Size of gap between two bony fragments after debridement was 10 cm. New bone filled up the resultant gap after extensive debridement following a distraction. There was hyporegenerate due to instability as the bone was grossly osteoporotic and was taken care of as mentioned above.

There was no recurrence of infection but pin-tract infection (PTI) was seen. PTI was classified as per Paley classification and managed accordingly. They were mostly type 1 and 2 PTIs. Two wires were substituted due to type 3 PTI and placed at different levels [7]. There was stiffness at knee joint (final range of motion was 15°–15°–120°). The ankle was fused in 10° and 5° of equinus and valgus deformity, respectively. The patient was happy with these results and more so with independent walking. Total duration of the ring was approximately 1 year 11 months.

[Calculation: (1) 20 cm shortening = 180 days (2 cm shortening persisted). (2) 10 cm regenerate = 100 days but half distraction days for 30 days. So 130 days. (3) AM 2 cycles = 40 days. (4) Twice the period for consolidation: $180 \times 2 = 360$ days. So, total $180 + 130 + 40 + 360 = 710$ days (1 year 11 months approximately. The regenerate got consolidated while the limb was undergoing external bone transport)].

Discussion

Problems with the case were gap to be created after debridement and existing shortening of the limb. Therefore, a long period of treatment with the apparatus was anticipated. This was discussed with the parents of the patient. The total duration of treatment was approximately 1 year 11 months. Ilizarov system is the most successful in this type of situation. Ilizarov concept and technology had indeed revolutionized

orthopedic treatment. It forms bone without bone grafting and facilitates soft-tissue closure without plastic reconstruction [8]. Neurofibromatosis may be a cause of CPT in 40–80% of cases [9]. Infection, repeated surgery leading to avascularity, and neurofibromatosis were three very strong prognostic factors that could influence the outcome in her treatment.

During debridement, we excised all unhealthy and dead tissues to ensure healthy stump ends and surrounding soft tissues. This tactic helped not only to ensure complete excision of neurofibromatosis elements but also prevented recurrence of infection. During the distraction, hyporegenerate created another problem. Paley enumerated likely causes of hyporegenerate and suggested means how to tackle each. He described technical factors such as traumatic corticotomy, initial diastasis, instability of apparatus, too rapid distraction, and patient factors such as infection, malnutrition, metabolic, and vascular as probable causes [7]. Augmentation of the apparatus, slowing down the distraction rate, employing AM, encouraging weight-bearing, and improving the nutrition of the patient helped us to tide over this problem. External bone transport [10] was started after docking was achieved to lengthen the entire limb.

Probably, frequent tightening of wires, augmentation of the apparatus, and ensuring meticulous pin-tract care helped us to have lesser PTIs.

The patient became anxious when hyporegenerate appeared and the process had to be slowed down. Uncertainty prevailed assuming an anticipated failure. It was a hard task to convince the patient and her parents that the phase would pass away and we would succeed. During the whole period of so many months, a vigil was ensured carefully to counsel at appropriate time of crisis. This paid the dividend and we could achieve our goal. It is of paramount importance that necessary steps are to be taken in maintaining the good mental health of the patient during such long period of confined treatment.

Conclusion

Chronic non-union associated with neurofibromatosis and infection is a very challenging situation in orthopedics. Radical debridement is the key to prevent recurrence of both conditions. Ilizarov system plays the most significant role in forming regenerate by distraction. Hyporegenerate and PTIs are to be tackled meticulously to prevent complications and attain the desired result. The patient could achieve painless unaided walking after the treatment. Probably instilling self-confidence in the patient was one of the most important aspects to sustain such an arduous journey.



Clinical Message

The greatest achievement, in this case, is that an amputable limb was salvaged. Return of function without residual infection added to the final outcome. This feat could be possible only with Ilizarov, a versatile modern orthopedic technology. We strongly believe that such technology should be studied and mastered in a country like India to reduce the osteoarticular burden of society.

References

1. Bhandari M, Fong K, Sprague S, Williams D, Petrisor B. Variability in the definition and perceived causes of delayed unions and non-unions: A cross-sectional, multinational survey of orthopaedic surgeons. *J Bone Joint Surg Am* 2012;94:e1091-6.
2. Nolte PA, van der Krans A, Patka P, Janssen IM, Ryaby JP, Albers GH. Low-intensity pulsed ultrasound in the treatment of nonunions. *J Trauma* 2001;51:693-703.
3. Zura R, Rocca GJ, Mehta S, Harrison A, Brodie C, Jones J, et al. Treatment of chronic (>1 year) fracture nonunion: Health rate in a cohort of 767 patients treated with low-intensity pulsed ultrasound (LIPUS). *Injury* 2015;46:2036-41.
4. Rosen H. Nonunion and malunion. In: Browner BD, Levine AM, Jupiter JB, editors. *Skeletal Trauma: Fractures, Dislocations, Ligamentous Injuries*. Philadelphia, PA: WB Saunders; 1998. p. 501-41.
5. Baruah RK, Baruah JP, Sunder SS. Acute shortening and re-lengthening (ASRL) in infected non-union of tibia advantages revisited. *Malaysian Orthop J* 2020;14:47-56.
6. Baruah RK, Patowary S. Accordion manoeuvre: A bloodless tool in Ilizarov. *J Limb Lengthen Reconstr* 2018;4:11-9.
7. Paley D. Problems, obstacles and complications of limb lengthening. In *Operative Principles of Ilizarov* By ASAMI group. Baltimore, Maryland, USA: Williams and Wilkins; 1991. p. 352-65.
8. Cattaneo R, Catagni M, Johnson E. The treatment of infected non unions and segmental defects of the tibia by the methods of Ilizarov. *Clin Orthop Relat Res* 1992;280:143-52.
9. Kulkarni RM. Congenital pseudoarthrosis of the tibia. In Kulkarni GS, editor. *Text Book of Orthopaedics and Trauma*. 2nd ed., Vol. 2. New Delhi: Jaypee Brothers Medical Publishers (P) Ltd.; 2008. p. 1674-80.
10. Golyakhovsky V, Frankel VH. Segmental bone transport in large bone loss and in severe infections. In Golyakhovsky V, Frankel VH, editors. *Text Book of Ilizarov Surgical Techniques. Bone correction and lengthening*. 1st ed. New Delhi: Jaypee Brothers Medical Publishers (P) Ltd.; 2010. p. 187-8.

Conflict of Interest: Nil

Source of Support: Nil

Consent: The authors confirm that informed consent was obtained from the patient for publication of this case report

How to Cite this Article

Baruah RK, Sunder SS. Success Story of Ilizarov in Chronic Non-union of Tibia with 18 Years of Morbidity: A Case Report. *Journal of Orthopaedic Case Reports* 2021 July;11(7): 12-15.