



Case report

Distraction osteogenesis at the site of previously cystic bone lesion of femur: A case report

Mujaddid Idulhaq, Asep Santoso^{*}, Ismail Mariyanto, Pamudji Utomo

Department of Orthopaedic and Traumatology, Universitas Sebelas Maret, Prof. Dr. R. Soeharso Orthopaedic Hospital, Surakarta, Indonesia

ARTICLE INFO

Keywords:

Distraction osteogenesis
Femur
Cystic bone lesion

ABSTRACT

Introduction and importance: Limb length discrepancy can be occurred as residual problems after bone cyst treatment. Distraction osteogenesis is one of surgical option to treat this problem.

Case presentation: A male 27 years old came with 3 cm residual lower limb discrepancy after treatment of femoral aneurismal bone cyst (ABC). Distraction osteogenesis has been successfully performed to treat this case at the site of previously cystic bone lesion.

Conclusion: Good quality of callus could be expected at the site of distraction osteogenesis that previously a cystic bone lesion.

1. Introduction

Benign bone cyst is one of common problem occurred in lower extremity bone [1,2]. Several techniques have been described to treat this problem [2,3]. Despite good bone healing after fixation, some residual problem happened to the patients. One of them is limb length discrepancy (LLD) [4]. This problem can be significantly altered clinical function if it is occurred on lower extremity. We report a case of successful distraction osteogenesis at the site of previously cystic bone lesion of femur to treat lower limb length discrepancy. Written informed consent is performed prior to this publication. The work has been reported in line with the SCARE criteria [5].

2. Case presentation

We reported a case of male 27 years old with chief complain of limping gait due to lower limb length discrepancy (LLD). He had history of pathologic fracture of right femur due to Aneurismal bone cyst (ABC) and treated with internal fixation and bone grafting at 1.5 year before. No complication occurred after the previous internal fixation surgery (Fig. 1A, B). The patient has been able to perform full weight-bearing walking without assistive device.

Physical examination of the right femur revealed an operation scar at lateral site of the femur region with no sign of infection. There was a full range of movement (ROM) of the right hip and knee joint. No

abnormality on contralateral side of lower leg. Lower leg length measurement showed a 3 cm LLD. The patient wanted to have an equal length of his lower leg. Patient has no other complications, drugs allergy, or adverse reactions related to prior surgery. After informed consent regarding the surgical procedure and possible complication, distraction osteogenesis was planned to the patient.

Surgery performed under spinal anesthesia in supine position without tourniquet. Surgery was performed by two surgeons (MI and AS). Incision was performed through previous incision with meticulous bleeding control. First, removal of the previous implant was performed. Distraction osteogenesis of the femur performed with the use of 40 cm monorail fixator (® B-fix, Aike, Shanghai Medical Instrument, China). Osteotomy site for bone lengthening was performed at the distal part of the previous cystic bone lesion (Fig. 2 A, B, C).

Postoperative management was given include cephalosporin antibiotic and pain control with ketorolac injection. Standard rehabilitation protocol was performed to the patients. Patient was allowed for early ROM exercise and non-weight bearing mobilization with crutch. Distraction osteogenesis was started at day 10th after surgery with 1 mm lengthening per-day. Post-Operative follow-up was performed at 2 weeks, 1 month, and every 1 months thereafter. During the follow-up, equal leg length was obtained at 1.5 month post-operatively. At 11 months post-operative the new bone formation has been remodeled with good bone quality and the patient could perform full-weight bearing walking without assistive device (Fig. 3 A, B). The patient experienced

^{*} Corresponding author at: Department of Orthopaedic and Traumatology, Universitas sebelas Maret, Prof. Dr.R. Soeharso Orthopaedic Hospital, Jl. Jenderal Ahmad Yani, Pabelan, Surakarta, Indonesia.

E-mail address: asepsantoso@staff.uns.ac.id (A. Santoso).

<https://doi.org/10.1016/j.ijscr.2021.106153>

Received 26 May 2021; Received in revised form 27 June 2021; Accepted 27 June 2021

Available online 29 June 2021

2210-2612/© 2021 Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

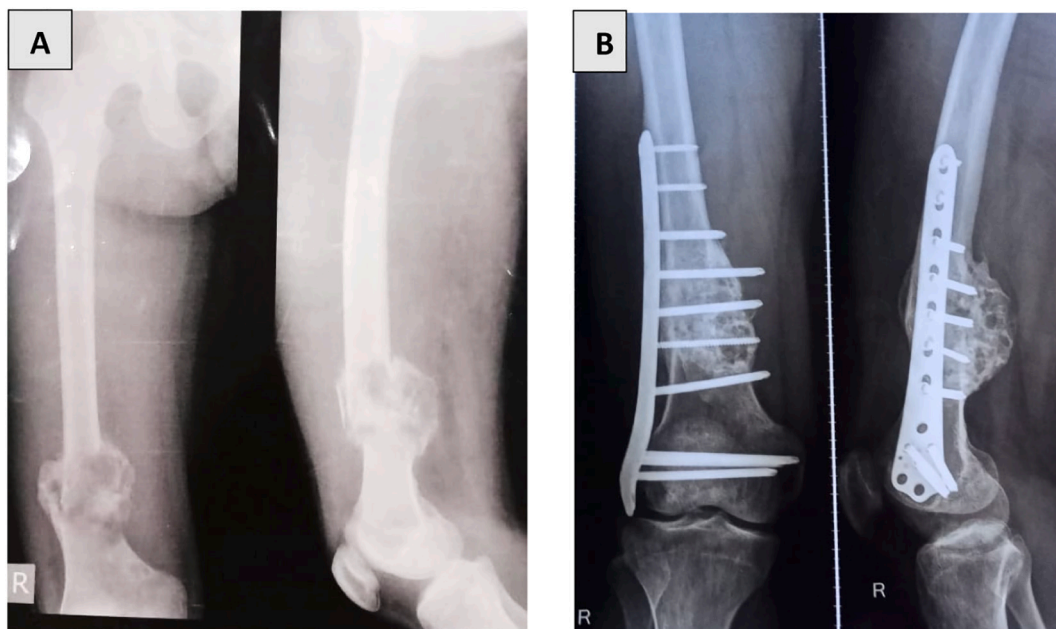


Fig. 1. A. Pathologic femur fracture due to Aneurismal bone cyst (ABC), B. At 1 year after internal fixation and bone grafting.

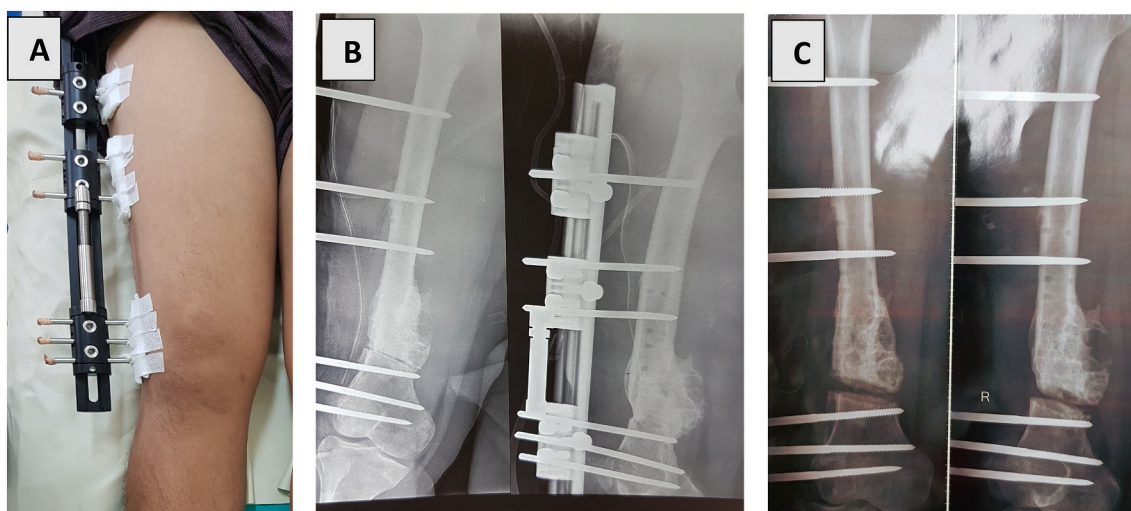


Fig. 2. A. Monorail Fixator, B. Immediate Post-operative radiograph after monorail application, C. After 2 weeks of bone lengthening.

knee joint stiffness post-operatively with 0-70° of flexion. At 1 year post-operatively, removal of the prosthesis in combination with surgical release were performed to increase the knee joint motion (Fig. 3 C, D). The final follow-up (3 years) examination the patients has full range of knee joint movement and satisfactory clinical outcome and no subjective complain.

3. Discussion

Several factors could affect the quality of bone regeneration in distraction osteogenesis. Local tissue factors such as the presence of bone pathology, previous internal fixation and soft tissue quality can alter bone regeneration in distraction osteogenesis [6,7]. Therefore, the presence of previous bone cyst in our recent case may also possible to affect callus formation during distraction osteogenesis. Furthermore, the presence of previous plate-screw internal fixation which needed removal off course may damage the soft tissue and vascularization to the bone. In our recent case, previous bony cyst site was located at the meta-

diaphyseal junction of the distal femur. We perform osteotomy for the distraction site at the most distal part of the previous cyst near the metaphyseal area of distal femur to increase the chance of new bone formation.

Hamanishi et al. reported several types of calluses in distraction osteogenesis include: external, straight, attenuated, opposite, pillar, and agenetic [7]. In our recent case, it was found that the callus is the external type. The external type of callus was the best type of callus in distraction osteogenesis which the dimension of the callus is beyond the size of previous bone diameter (Fig. 3B, C). We also found that the remodeling process resulted a higher bone density compared the previous condition prior to distraction process. This case showed that good bone formation can be expected at the site of previous bone cyst.

Several previous literatures presented distraction osteogenesis in bone cyst cases. Verdiyev et al. in a 30 years review reported a total 25 cases of bone cyst (20 uni-cameral and 5 ABCs) which treated with distraction osteogenesis [1]. Among them, complete bone healing was found in 15 cases, nine case had residual bone lucency, and one case had

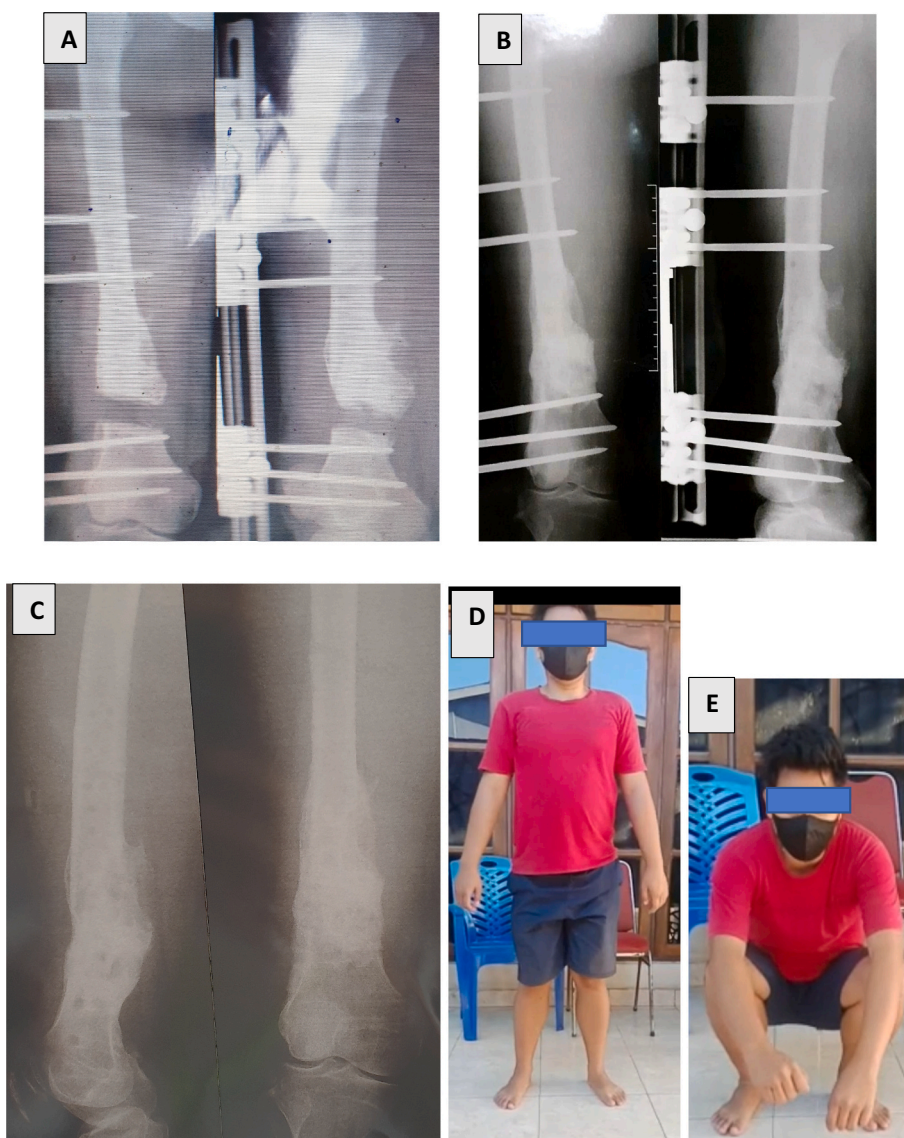


Fig. 3. A. At 1 month of bone lengthening, B. At 11 months after surgery, C. After removal of monorail fixator, D and E. Clinical picture at final follow-up (3 year).

poor outcome due to recurrency of the cyst. Another report by Acan et al., they reported a case of pediatric humerus aneurismal bone cyst which treated with intra-medullary bone lengthening [8]. They successfully perform 5 cm lengthening of the humeral bone.

4. Conclusion

Distraction osteogenesis is an effective procedure to manage residual problem after cystic bone lesion. Good quality of callus could be expected at the site of distraction osteogenesis that previously a cystic bone lesion.

Declaration of competing interest

None to declare.

Acknowledgments

None.

References

- [1] V. Verdiyev, F. Verdiyev, Application of distraction osteogenesis in managing bone cysts, *J. Orthop. Traumatol.* 15 (2014) 103–109, <https://doi.org/10.1007/s10195-013-0272-9>.
- [2] A. Roposch, V. Saraph, W.E. Linhart, Flexible intramedullary nailing for the treatment of unicameral bone cysts in long bones, *J. Bone Joint Surg. Am.* 82 (2000) 1447–1453, <https://doi.org/10.2106/00004623-200010000-00011>.
- [3] L. Eralp, F.E. Bilen, S.R. Rozbruch, M. Kocaoglu, A.I. Hammoudi, External fixation reconstruction of the residual problems of benign bone tumours, *Strateg. Trauma Limb Reconstr.* 11 (2016) 37–49, <https://doi.org/10.1007/s11751-016-0244-8>.
- [4] H.S. Kim, K.S. Lim, S.W. Seo, S.P. Jang, J.S. Shim JS, Recurrence of a unicameral bone cyst in the femoral diaphysis, *Clin. Orthop. Surg.* 8 (2016) 484–488, <https://doi.org/10.4055/cios.2016.8.4.484>.
- [5] for the SCARE Group, R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, The SCARE 2020 guideline: updating consensus Surgical CASE REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.
- [6] G.A. Hosny, Limb lengthening history, evolution, complications and current concepts, *J. Orthop. Traumatol.* 21 (2020) 3, <https://doi.org/10.1186/s10195-019-0541-3>.
- [7] C. Hamanishi, Y. Yasuwaki, H. Kikuchi, S. Tanaka, K. Tamura, Classification of the callus in limb lengthening radiographic study of 35 limbs, *Acta Orthop. Scand.* 63 (1992) 430–433.
- [8] A.E. Acan, O. Basci, H. Havitcioglu, Aneurysmal bone cyst healing response with intramedullary lengthening nail, *Acta Orthop. Traumatol. Turc.* 52 (2018) 232–235, <https://doi.org/10.1016/j.aott.2017.04.005>.