

A Commentary on “Clinical and Functional Outcomes of Ulnar Lengthening in the Treatment of Masada Type I Forearm Deformities in Hereditary Multiple Osteochondromas”

Alireza Moharrami^{1,*}, Hamid Arabi²

¹ Resident, Department of Orthopedic Surgery, Joint Reconstruction Research Center, Tehran University of Medical Sciences, Tehran, Iran

² Orthopedic Surgeon, Joint Reconstruction Research Center, Tehran University of Medical Sciences, Tehran, Iran

*Corresponding author: Alireza Moharrami; Department of Orthopedic Surgery, Joint Reconstruction Research Center, Tehran University of Medical Sciences, Tehran, Iran. Tel: +98-9122432939, Email: a.moharrami@gmail.com

Received: 12 January 2020; Revised: 17 February 2020; Accepted: 28 February 2020

Keywords: Bone Lengthening; Hereditary Multiple Exostosis; Multiple Osteochondromas

Citation: Moharrami A, Arabi H. A Commentary on “Clinical and Functional Outcomes of Ulnar Lengthening in the Treatment of Masada Type I Forearm Deformities in Hereditary Multiple Osteochondromas”. *J Orthop Spine Trauma* 2020; 6(1): 24-5.



Background

Hereditary multiple exostoses (HME) is a benign condition that begins close to the growth plate and can develop several deformities such as metaphyseal remodeling, bony deformities, and asymmetric long bone growth (1). HME could involve the forearm as a common deformity in 60% of patients (2) and may cause ulnar shortening as a result of premature closure of the epiphyseal growth plate. The severity of the disease has a correlation with the severity of the deformities and the patients suffer from cosmetic and functional defects (3, 4). Many studies reported that the deformity could cause restricted range of motion (ROM), supination, and pronation (5, 6). Recently, various treatments have been proposed to correct such deformities in the forearm, including ulnar lengthening and radial shortening. However, no specific protocol can be found in the literature regarding overall management and surgical options (1). Previous case reports have investigated ulnar lengthening as a surgical method in treatment of HME-induced forearm deformities.

There were several studies regarding ulnar lengthening in HME patients with forearm deformity. In 1986, Pritchett reported excellent results for ulnar lengthening in such patients (7). In 2006, a study by Matsubara et al. on seven HME patients with forearm deformity revealed that gradual ulnar lengthening with excision of osteochondromas had satisfactory results in all patients (8). Similarly, Cho and Jung reached the same results in 4 cases of forearm deformity that were corrected with the Ilizarov method (5). Tang et al. demonstrated that a combination of ulnar lengthening and osteochondroma excision in HME patients with Masada type I forearm deformity and ulnar shortening more than 15 mm had satisfactory outcomes (9). Litzelmann et al. suggested radial head dislocation as the indication of ulnar lengthening in patients with HME, and also, recommended performing radial osteotomy near to the end of the growth plate (1). Recent studies reached similar results in the effectiveness of ulnar lengthening in patients with HME (10-12). Ahmed reported that gradual ulnar lengthening without tumor excision was effective in HME patients with Masada type IIb forearm deformity (10).

Here, we discuss the new article by Baghdadi et al.

published in the journal of Hand Surgery (13). They evaluated the outcomes of ulnar lengthening in Masada type I forearm deformity in patients with HME and found good outcomes. The authors stated that the main benefit of forearm reconstruction in Masada type I was preventing dislocation of the radial head, which results in substantial functional disability.

In these patients, the most commonly practiced surgical treatments are excision of the osteochondromas and ulnar lengthening. Previous studies have shown clinical and radiographic improvements after these treatments (1, 11, 14), but whether the changes in radiographic measurements are correlated with patient-reported functional improvements remains unknown.

Discussion

The study by Baghdadi et al. showed a significant clinical and radiographic improvement, but only a modest improvement in the Disabilities of the Arm, Shoulder, and Hand (DASH) score and mental health scales of the 36-Item Short-Form Survey (SF-36) for quality-of-life, and no improvement in physical health scales of the SF-36 scoring system (13). Here, we would like to propose some suggestions for future studies of this sort.

First, in the method of treatment, all of the patients in the study by Baghdadi et al. (13) had Masada type I forearm deformity and were treated with gradual lengthening of the forearm using an external fixator, and a concomitant excision of the ulnar osteochondroma in seven patients. The excision of ulnar osteochondroma could increase the risk of neurovascular impairment in the early phases. It is better to treat all patients with the same method and device, before comparing the clinical and functional outcome.

Second, there are conflicting results on the functional outcome of ulnar lengthening in previous studies (1, 11, 14). In the study by Baghdadi et al., the significant improvement in the forearm movement was only observed in supination. There were no significant changes in flexion, extension, and pronation during the study period (13). Maybe the discrepancies in the reports are due to small sample size that usually limits many studies in this field. Therefore, to get a definite conclusion, we need studies

with larger sample sizes or meta-analysis studies.

Third, the ulnar overlengthening may have beneficial effects on the radial bowing, theoretically. In the study by Baghdadi et al., the radiographic indices showed improvement of the ulnar shortening by a mean of 11.3 mm at the last follow-up (13). Some previous studies performed radial osteotomy at the end of treatment to achieve better outcomes. This study proved that not only we need radial osteotomy, but also we can use ulnar overlengthening to achieve better outcomes with fewer complications.

Finally, Baghdadi et al. had no major complications during the study. They performed forearm lengthening with an intramedullary pin which may prevent ulnar deviation (13). Previous studies reported that using intramedullary pinning could reduce time to union, non-union, and subsequent fractures (15, 16). Based on the study by Baghdadi et al., it is recommended to use intramedullary pinning during ulnar lengthening in HME cases (13). However, further studies with larger sample sizes are needed.

There are acceptable functional and radiological outcomes with ulnar lengthening technique in forearm deformity. Baghdadi et al. revealed that this technique had good outcomes with Masada type I forearm deformity, and although they did not perform any radial osteotomy, they had better functional outcomes in comparison with previous studies (13). Ulnar lengthening with an external fixator and intramedullary pinning without radial osteotomy may be a beneficial method with the acceptable outcome for Masada type I forearm deformity.

Conflict of Interest

The authors declare no conflict of interest in this study.

Acknowledgments

None.

References

- Litzelmann E, Mazda K, Jehanno P, Brasher C, Pennecot GF, Ilharreborde B. Forearm deformities in hereditary multiple exostosis: Clinical and functional results at maturity. *J Pediatr Orthop*. 2012;32(8):835-41. doi: [10.1097/BPO.0b013e3182694e40](https://doi.org/10.1097/BPO.0b013e3182694e40). [PubMed: 23147628].
- Shapiro F, Simon S, Glimcher MJ. Hereditary multiple exostoses. Anthropometric, roentgenographic, and clinical aspects. *J Bone Joint Surg Am*. 1979;61(6A):815-24. [PubMed: 225330].
- Massobrio M, Antonietti G, Pellicano G, Necci F. Single forearm radius and ulna asymmetric lengthening in multiple cartilaginous exostoses: A case report and a long-term follow-up. *J Pediatr Orthop B*. 2015;24(6):561-6. doi: [10.1097/BPB.0000000000000212](https://doi.org/10.1097/BPB.0000000000000212). [PubMed: 26196368].
- Noonan KJ, Levenda A, Snead J, Feinberg JR, Mih A. Evaluation of the forearm in untreated adult subjects with multiple hereditary osteochondromatosis. *J Bone Joint Surg Am*. 2002;84(3):397-403. doi: [10.2106/00004623-200203000-00010](https://doi.org/10.2106/00004623-200203000-00010). [PubMed: 11886909].
- Cho YJ, Jung ST. Gradual lengthening of the ulna in patients with multiple hereditary exostoses with a dislocated radial head. *Yonsei Med J*. 2014;55(1):178-84. doi: [10.3349/ymj.2014.55.1.178](https://doi.org/10.3349/ymj.2014.55.1.178). [PubMed: 24339304]. [PubMed Central: PMC3874895].
- Clement ND, Porter DE. Forearm deformity in patients with hereditary multiple exostoses: Factors associated with range of motion and radial head dislocation. *J Bone Joint Surg Am*. 2013;95(17):1586-92. doi: [10.2106/JBJS.L.00736](https://doi.org/10.2106/JBJS.L.00736). [PubMed: 24005199].
- Pritchett JW. Lengthening the ulna in patients with hereditary multiple exostoses. *J Bone Joint Surg Br*. 1986;68(4):561-5. [PubMed: 3488318].
- Matsubara H, Tsuchiya H, Sakurakichi K, Yamashiro T, Watanabe K, Tomita K. Correction and lengthening for deformities of the forearm in multiple cartilaginous exostoses. *J Orthop Sci*. 2006;11(5):459-66. doi: [10.1007/s00776-006-1047-4](https://doi.org/10.1007/s00776-006-1047-4). [PubMed: 17013733].
- Tang ZW, Cao YL, Liu T, Chen T, Zhang XS. Management of forearm deformities with ulnar shortening more than 15 mm caused by hereditary multiple osteochondromas. *Eur J Orthop Surg Traumatol*. 2013;23(5):611-8. doi: [10.1007/s00590-012-1033-9](https://doi.org/10.1007/s00590-012-1033-9). [PubMed: 23412166].
- Ahmed AARY. Gradual ulnar lengthening by an Ilizarov ring fixator for correction of Masada IIb forearm deformity without tumor excision in hereditary multiple exostosis: Preliminary results. *J Pediatr Orthop B*. 2019;28(1):67-72. doi: [10.1097/BPB.0000000000000514](https://doi.org/10.1097/BPB.0000000000000514). [PubMed: 29995654].
- D'Ambrosi R, Barbato A, Caldarini C, Biancardi E, Facchini RM. Gradual ulnar lengthening in children with multiple exostoses and radial head dislocation: Results at skeletal maturity. *J Child Orthop*. 2016;10(2):127-33. doi: [10.1007/s11832-016-0718-8](https://doi.org/10.1007/s11832-016-0718-8). [PubMed: 26910403]. [PubMed Center: PMC4837170].
- Li Y, Han B, Tang J, Chen M, Wang Z. Identification of risk factors affecting bone formation in gradual ulnar lengthening in children with hereditary multiple exostoses: A retrospective study. *Medicine (Baltimore)*. 2019;98(5):e14280. doi: [10.1097/MD.00000000000014280](https://doi.org/10.1097/MD.00000000000014280). [PubMed: 30702592]. [PubMed Central: PMC6380801].
- Baghdadi S, Arabi H, Farhoud A, Moharrami A, Baghdadi T. Clinical and functional outcomes of ulnar lengthening in the treatment of masada type i forearm deformities in hereditary multiple osteochondromas. *J Hand Surg Am*. 2020;45(9):876-. doi: [10.1016/j.jhsa.2020.02.010](https://doi.org/10.1016/j.jhsa.2020.02.010). [PubMed: 32253060].
- Refsland S, Kozin SH, Zlotolow DA. Ulnar distraction osteogenesis in the treatment of forearm deformities in children with multiple hereditary exostoses. *J Hand Surg Am*. 2016;41(9):888-95. doi: [10.1016/j.jhsa.2016.06.008](https://doi.org/10.1016/j.jhsa.2016.06.008). [PubMed: 27491595].
- Jager T, Popkov D, Lascombes P, Popkov A, Journeau P. Elastic intramedullary nailing as a complement to Ilizarov's method for forearm lengthening: A comparative pediatric prospective study. *Orthop Traumatol Surg Res*. 2012;98(4):376-82. doi: [10.1016/j.otsr.2012.01.007](https://doi.org/10.1016/j.otsr.2012.01.007). [PubMed: 22560591].
- Launay F, Jouve JL, Viehweger E, Guillaume JM, Jacquemier M, Bollini G. Progressive forearm lengthening with an intramedullary guidewire in children: Report of 10 cases. *J Pediatr Orthop*. 2004;24(1):21-5. [PubMed: 14676529].