REVIEW ARTICLE

Humerus Lengthening with a Motorized Intramedullary Nail: A Systematic Review of Outcomes and Complications

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ABSTRACT

Aim: In the past decade, internal limb lengthening nails have gained popularity. In this study, we aim to systematically review the literature on humerus limb lengthening with a motorized intramedullary nail (MIN). We intend to assess the outcome and complications of this technique. **Materials and methods:** A systematic review was performed in the following databases: PubMed, Embase, Web of Science, and the *Journal of Limb Lengthening and Reconstruction*. The inclusion criteria included limb lengthening of the humerus using an intramedullary nail, clinical studies, all levels of evidence, and no restriction to the date of publication.

Results: Our search yielded 239 journal articles. A total of nine articles remained relevant based on the inclusion and exclusion criteria. The total number of patients was 20, with 22 segments lengthened. The mean age of the patients was 20.8-year-old [standard deviation (SD), 12.0; range, 13–51]. The mean gained length was 5.7 cm (SD, 0.9; range, 5–7.5) with a mean distraction protocol of 0.82 mm/day (SD, 0.2; range, 0.6–1). The average duration of lengthening was 71.6 days (SD, 12.8; range, 50–93), and the mean duration of consolidation was 192.3 days (SD, 40.5; range, 120–228). Reported complications included a range of motion (ROM) limitation, hardware failure, and hypertrophic bone regeneration. **Conclusion:** Humeral lengthening with an MIN provides favourable outcomes with low complication rates. Future high-level studies should focus on comparing long-term outcomes of humeral lengthening utilising internal and external fixation techniques.

Clinical significance: Humeral lengthening using MIN can be used safely. Each surgical approach and type of nail have different risks and benefits. These should be carefully discussed when planning the surgery.

Keywords: Clinical outcomes, Humerus, Limb lengthening, Motorized intramedullary nail, Systematic review.

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INTRODUCTION

Humeral length discrepancy has an important impact on a functional and aesthetic point of view. Patients may suffer from impaired activities of daily living, difficulty performing in sports or decreased self-image.¹ It results from congenital or acquired lesions such as multiple exostosis, osteomyelitis, trauma, unicameral bone cysts or premature growth disturbance.^{2–4} A common reason for lengthening in unilateral cases is shortening above 5 cm and angulation above 20°.⁵

Discrepancies in humeral length were originally treated with the Ilizarov circular frames, monolateral fixators and hexapod circular frames.⁶⁻⁹ However, external devices have been limited by their complications including soft tissue tethering, pin site infections, neurovascular entrapment and joint contracture.^{1,5,8,10-12} In the past decade, lengthening with an MIN has gained popularity.¹³⁻¹⁵ It was initially introduced for femoral and tibial lengthening given their bigger medullary canal, and later adapted for the humerus.

Two MINs that are commonly used are the PRECICE nail (Nuvasive, CA, USA)¹⁶ and the FITBONE nail (WITTENSTEIN Intens GmbH, Igersheim, Germany).¹⁷ The PRECICE nail mechanism is based on an electromagnetic field. After the surgery, the patients are given an external remote control that is used to rotate the magnet inside the intramedullary nail. These magnets rotate the gears, which turns a drive screw that then extend the telescopic nail. Regarding the FITBONE mechanism, it requires the placement of a motor antenna in the subcutaneous tissue. A device is used to transmit, by induction, electric current to the antenna that then go through the wire to reach the motor that rotate the spindle and elongates the nail. It was hypothesised that, compared to external fixator, there

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would be less pin site infections, lesser soft-tissue damage and pain, better joint movement, and more patient comfort with the MIN.^{16–21}

In this study, we aim to systematically review the literature on humeral lengthening with an MIN. We intend to assess the outcomes and complications of this technique. We hypothesise that overall outcomes are favourable with regard to amount of length gained, and the rate of substantial complications is low.

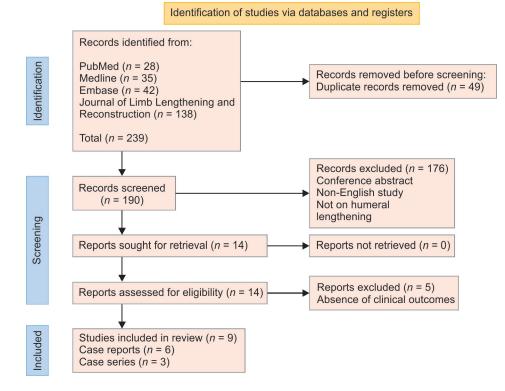
MATERIALS AND METHODS

Search Strategy

A systematic review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)

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Flowchart 1: PRISMA diagram of the included studies. Flowchart illustrating the inclusion of the studies with the steps involved in the screening process. The separation highlighted in blue on the left shows the different sections of the process.



guidelines.²² The search was performed in the following databases: PubMed, Embase (*via* Ovid), Web of Science (*via* Ovid) and in the *Journal of Limb Lengthening and Reconstruction*. All databases were searched from their start to 14 June 2022. The following keywords were used in the search strategy: Humerus and lengthening and intramedullary. Synonyms of these terms were also used but not listed here. Each reference section was hand screened in order to identify additional studies. Endnote X9 (Clarivate, Camelot UK Bidco Limited) was used to remove the duplicates and tract any removal or addition of study.

Inclusion and Exclusion Criteria

The following inclusion criteria were used in our systematic review: Limb lengthening of the humerus using an MIN, clinical studies, all level of evidence and no restriction to date of publication.

Studies were excluded if they met any of the following criteria: Non-English articles, lengthening of the lower limbs and articles published in abstract form only. In addition, articles about limb lengthening that did not differentiate patients with humerus lengthening from the other limbs were excluded. This was because extraction of data specific to the humerus was not possible, and multiple attempts were made to contact the authors of these articles to get specific results of those patients, but the response and collaboration was extremely poor.

Data Collection/Extraction

Two authors (JPL and NA) screened the titles and abstracts of the included articles independently. When any discrepancy between reviewers arose, it was resolved by discussion between them and if necessary, with other members of the research team (Y.M and A.A). Two authors (JPL and NA) then independently retrieved data from the included studies in Microsoft Excel 2013 [Microsoft,

Redmond, Washington, United States of America (USA)]. The information was categorised into basic article information (e.g. title, authors, year of publication, journal, and country), patient background information and methodology details (e.g. sample size, sex, age, preoperative assessment, inclusion/exclusion criteria and indication for surgery), surgical technique (e.g. segment lengthened and type of intramedullary nail) and post-operative outcomes and complications (e.g. duration of follow-up, end lengthening achieved, lengthening period, consolidation index, rate of distraction and post-operative ROM).

Statistical Analysis

The International Business Machines (IBM) Statistical Package for the Social Sciences (SPSS) Statistics, version 23.0, software (IBM, Armonk, New York, USA) was used to analyse the data. This included a descriptive analysis of all variables, including frequencies, percentages, means, SDs, and other basic statistics.

Results

Our search yielded 239 titles (Flowchart 1). After removing duplicates, 190 articles were screened based on titles and abstracts. No new articles were found after reviewing the bibliography of each paper. A total of nine articles remained relevant based on the inclusion and exclusion criteria. Every paper was published between 2016 and 2021.

The total number of patients was 20, with 22 segments lengthened (Table 1). 45% of segments involved the left humerus and 55% the right. The male to female ratio was 1.2:1, and the mean age of the patients was 20.8 years (SD, 12.0; range, 13–51). The mean preoperative shortening of the humerus was 6.0 cm (SD, 1.6; range, 4–9). More than six specific etiologies for the shortening

Table 1: Preope	rative inforr	mation of th	Table 1: Preoperative information of the included studies						
Study	Patients (n)	Segments (n)	Indication for lengthening (n indication)	Proportion (male:female)	Proportion (left:right)	Mean age (year)	Mean age Associated deformities (year) (n deformity)	Humerus length discrepancy (cm shorter)	Type of nail used
Acan et al. ³⁴	-	1	Recurrent aggressive aneurysmal bone cysts	1:0	1:0	13	Deformity noted but not described	5	PRECICE
Fürmetz et al. ²³	4	4	 2 Erb–Duchenne-type obstetric palsy 1 traumatic growth arrest in childhood 1 post-traumatic shortening after complex fracture and non-union 	3:1	3:1	29	Nil described	5.5	3 FITBONE 2 PRECICE
Ghafoor et al. ³⁵	-	-	Traumatic upper plexus palsy	0:1	0:1	13	Nil described	Ŋ	PRECICE
Hammouda et al. ¹⁰	Ŋ	Q	 multiple hereditary exostosis bone cyst/growth arrest growth arrest shoulder growth arrest 	2:3	3:3	20	1 45°-external torsion 1 15° varus, 13°-apex anterior 1 10° varus, 12°-apex anterior	5.1	PRECICE
Hoel et al. ³⁶	-	-	Traumatic fracture at age 8 through a previously asymptomatic proximal right humeral cyst	0:1	0:1	14	Nil described	8.1	PRECICE
Kurtz et al. ¹¹	-	-	Bone cyst	0:1	0:1	15	Midshaft, oblique plane deformity composed of 15° varus and 13°-apex anterior	7.5	PRECICE
Morrison et al. ¹²	Ŋ	Q	1 septic physeal arrest 2 traumatic physeal arrest 1 brachial plexus palsy 1 unicameral bone cyst	4:1	2:4	15	2 with angular deformities, corrected at the corticotomy site	7	PRECICE
Tiefenboeck et al. ³⁷	-	-	Congenital shortening of the left humerus	0:1	1:0	32	Nil described	S	PRECICE
Zak et al. ³²	-	1	Humeral epiphyseal fracture in the childhood	1:0	0:1	38	Nil described	6.5	PRECICE

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were described. Among them, the most frequent were bone cyst (four), complex fractures (six), brachial plexus palsy (four) and septic growth arrest (two). Seven patients had associated deformities on exam and/or imaging. The intramedullary nail used in the majority of patients was PRECICE (20/22 lengthening).

Table 2 demonstrates the intra- and post-operative data obtained from the study. Most studies used an antegrade approach for nail insertion. The mean gained length was 5.7 cm (SD, 0.9; range 5–7.5) with a mean distraction protocol of 0.82 cm/day (SD, 0.2; range, 0.6–1). The average duration of lengthening was 71.6 days (SD, 12.8; range, 50–93). Mean duration of consolidation was 192.3 days (SD, 40.5; range, 120–228) with a respective consolidation index of 34.0 days/cm (SD, 7.1; range, 25–45).

As represented in Table 2, ROM limitations were variable across studies. Five studies had similar preoperative ROM and post-operative ROM. Fürmetz et al.²³ recorded a loss of ROM in every patient. One patient with the FITBONE nail had reduction of shoulder abduction by 20° and another had a reduction in shoulder abduction by 20° in addition to a reduction of shoulder flexion by 10°. One patient with the PRECICE nail had a 5° decreased in elbow extension with a shoulder abduction and flexion increased by 10°. The other studies did not quantify the loss in ROM. Among them, described changes included a minimal loss of shoulder hyperabduction and a temporary flexion contracture of elbow. Hammouda et al.¹⁰ described the case of a patient with complex regional pain syndrome that sustained a significant reduction in shoulder flexion/extension postoperatively, without quantification of the limitation. No functional limitations were reported. Nevertheless, certain complications required modifications to the treatment protocol. One patient in the FITBONE group experienced pain and irritation caused by the cord penetrating the rotator cuff. The MIN receiver and cord were removed at the end of the lengthening and the pain was cleared. In addition, a different patient in the FITBONE group suffered from flexion contractures of the elbow after the lengthening. He required Botulinum toxin injection and a Z-plasty of the bicep tendon to resolve this complication. Premature arrest of a FITBONE lengthening was required in one patient due to proximal migration of the humeral head. An obstacle noticed in the PRECICE group was limitation to ROM of the shoulder, which was resolved with physiotherapy and by decreasing the lengthening rate. Two patients from the PRECICE group suffered from a hardware breakage and had to be revised. One of them was due to a loss of fixation of the MIN and the other was caused by a PRECICE P2 crown failure. Finally, hypertrophic bone regenerate was seen in one patient in the PRECICE group. This complication was resolved by temporarily increasing the lengthening rate to 1.5 mm/day.

CONCLUSION

Humeral lengthening using MIN provides favourable outcomes with low complication rates. The future high-level studies should focus on comparing long-term outcomes of humeral lengthening utilising internal and external fixation methods.

Clinical Significance

This systematic review of 22 segments reveals that MIN humeral lengthening techniques can result in substantial length gain with high rate of success. Overall, the rate of complications was low for humerus limb lengthening; however, the treating surgeon should

be experienced in managing minor problems and obstacles to avoid increasing the rate of serious complications and their consequences.

In this review, the PRECICE intramedullary nail was significantly more used than the FITBONE. No study reported on the reasoning behind the choice of MIN. A major advantage is the size of the nails, where the PRECICE MIN has a diameter of 8.5–12.5 mm and FITBONE MIN has a diameter of 11–13 mm.

During the lengthening, multiple studies reported a temporary loss of shoulder ROM which did not require any intervention. While no ROM data were available for these studies, most of them were due to contractures of the surrounding muscles. However, most studies used a proximal/antegrade approach for nail insertion. It is described in the literature that an antegrade approach for MIN insertion in patients with humeral fracture can lead to a disruption of the rotator cuff and subsequent limitations in ROM.^{24,25} On the other hand, it was also reported that retrograde approach could lead to elbow stiffness in fracture treatment.²⁶ We found only one case of chronic elbow stiffness, which was treated using a retrograde approach.²³ Early physiotherapy during lengthening was suggested to prevent long-term limitation of the upper extremities.²³

The Ilizarov method and the monolateral fixators were shown to have a mean consolidation index of 27–32 days/cm^{27,28} and 24–32 days/cm, respectively.¹⁰ In this systematic review, we demonstrated that the mean consolidation index for intramedullary devices is 34 days/cm. This is coherent with a comparison that showed a similar consolidation index between magnetic internal fixation nails and external fixators.²⁹ Moreover, the results of this systematic review show a similar mean distraction protocol for MIN lengthening and external fixators. Both were shown to be effective with a distraction of 1mm/day, divided by 0.25 mm 4 times per day.^{29–31}

One of the main limitations of the MIN is its maximum length. Ring external fixators were shown to have a mean increase in humeral length of range 5–11.1 cm.^{1,5,9} Regarding the MIN, a mean humeral lengthening of 5.7 cm (5.0-7.5 cm) was obtained. In our systematic review, only 54% (10/22) of segments reached the targeted length gain. Among them, only two had a preoperative shortening of 50 mm, and the rest had more than 65 mm of shortening. The PRECICE nail lengthening capacity (i.e. stroke distance) is 50 mm for nails measuring less than 245 mm in length, and 80 mm for nails measuring more than 245 mm in length. On the other hand, the FITBONE nail has a maximum lengthening of 80 mm. To remediate this issue, most studies adjusted the acute distraction during the initial osteotomy to comply with the specifications of the MIN. Only one study reported a novel approach consisting of unlocking, backwinding and interlocking the telescopic nail to achieve a total gained length of 6.5 cm.³²

In a recent systematic review of motorized lengthening nails used for upper and lower extremities, MIN was found to have a 34% rate of complication when combining all severity scales.¹³ This is similar to our study which demonstrated a complication rate of 27% (6 over 22 segments). However, this is less in patients treated with external fixators. Comparison studies showed an MIN to external fixator complication ratio of 0.5–0.8:1.^{12,29}

As hypothesised, the risk of infection with intramedullary nails is significantly less than with external devices. In the latter, superficial infection was reported in up to 100% of cases, but the incidence of deep infection was much lower.³³ Among the included studies in this systematic review, infection was not seen in any patient. Other systematic reviews reported an infection rate of 0.8% with MIN lengthening in general (not specific to the humerus).¹³ This low

lable 2: Intra-	Iable 2: Intra- and post-operative information of the included studies Approach Duration of Duration until Ave. (anteorade vs lengthening consolidation consol	Unvernmation Duration of lenathenina	Duration until Consolidation	Average	Distraction protocol	Gained	Post-operative ROM	Reported complication (n-tvpe of	Duration of
Study	retrograde)	(days)	(days)	index (days/cm)	(mm/day)	lengths (cm)	(n-type of nail: ROM changes)	nail: complication)	follow-up
Acan et al. ³⁴	Antegrade	50	120	Not reported	1	5	Minimal loss shoulder hyperabduction	None	1 year
Fürmetz et al. ²³	3 antegrade and 1 retrograde	20	235	33.56	0.72	Full lengthening in 1 patient Average 5.5	1 FITBONE- reduction shoulder abduction from 20° 1 FITBONE-shoulder abduction decreased 20° and flexion decreased 10° 1 FITBONE-improved elbow extension 10° (with Botox injection and a Z-plastic). 1 PRECICE-5° decreased elbow extension, Shoulder abduction and flexion increased 10°	1 FITBONE: irritation and pain caused by the cord, penetrating the rotator cuff 1 FITBONE-reduction in elbow extension led to Botox injection and a Z-plastic of bicep tendon 1 PRECICE-failure after 10-mm lengthening due to breakage of the crown (PRECICE P2). New nail was inserted, and full lengthening was achieved without complications	Minimum follow-up of 6 months after nail removal
Ghafoor et al. ³⁵	Antegrade	77	Not yet achieved	Not reported	Not reported	5.2	Similar to preoperative	None	Not reported
Hammouda et al. ¹⁰	Antegrade	78	180	36	0.7	All segments achieved full lengthening Average 5.1	All patients maintained their preoperative ROM 1 patient (with complex regional pain syndrome)- significant reduction of post-operative shoulder flexion/extension	None	1.8 years (0.9–2.4 years)
Hoel et al. ³⁶	Retrograde	77	224	Not reported	1 switched to 1.5 at 7 weeks	7.5	Similar to preoperative	None	Not reported
Kurtz et al. ¹¹	Antegrade	63	210	Not reported	1 switched to 0.75 at 4 weeks	5	Similar to preoperative	None	Not reported
Morrison et al. ¹²	Retrograde and antegrade Distribution not reported	93	Not reported	Not reported	0.62	Full lengthening in 1 patient Average 5.7	1 PRECICE-temporary flexion contracture of elbow	1 PRECICE-loss of fixation of nail 1 PRECICE-non-compliance resulting in slow distraction	>6 months from lengthening
Tiefenboeck et al. ³⁷	Antegrade	Not reported	150		-	Ŀ	Similar to preoperative	None	Not reported
Zak et al. ³²	Antegrade	65	227	32.4	0.82	6.5	Similar to preoperative	None	15.5 months

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rate of infection is due to the lack of communication between the lengthening device and the exterior environment.

Several limitations exist in the current study. The main limitation is the low number of studies and segments. Despite our rigorous systematic review, only 22 segments were eligible for the analysis. Moreover, the studies have a low level of evidence (case reports or case series only). This is explained by the novelty of this technology, where all the included studies were published between 2016 and 2021. Studies with a larger number of patients are needed to improve the evidence on this topic. In addition, reported outcomes varied between the included studies, with some studies missing important outcomes such as maturation/consolidation index, functional scores or ROM. This makes outcome comparison difficult to interpret.

Availability of Data and Materials

Template data collection forms and extracted data are available upon request to the corresponding author.

AUTHORS' CONTRIBUTIONS

JPL: Critical review of study proposal, data collection, data analysis and manuscript preparation; NA: Writing assistance and data collection; YM: Critical review of the manuscript; AA: General advice, guidance and supervision; RCH: General advice, guidance and supervision; MB: Critical review of study proposal, design, methods; review of the manuscript, research and assistance.

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