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Humeral lengthening: Systematic Review and Meta Analysis

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Abstract

Background: The humerus lengthening was mostly regarded an aesthetic procedure. Recent study indicates, however, that the objectives of bilateral humeral lengthening in achondroplasia are not just cosmetic, but also to restore proportions between the upper and lower limbs, extend reach, and enhance the capacity to perform perineal personal hygiene. This study's objective was to conduct a comprehensive evaluation and analysis of the outcomes of humeral lengthening. Methods: During this systematic review and meta-analysis preparation, we adhered to the PRISMA statement criteria and followed the Cochrane handbook for systematic reviews of interventions. Results: We identified eight studies discussing humeral lengthening including a total number of 158 humeri in 129 patients. The age range of patients across the studies ranged between 6.5 and 20 years. The male gender was common and ranging from 33% to 78%. The humeral lengthening was conducted among 158 humeri among them 34 in the right side. The mean length achieved was 7.7 cm among the included eight145 studies ranging from 5.1 cm to 9 cm. Pooled analysis of the involved trials showed significant association between length pre and post humeral lengthening (OR = -6.08; 95% CI: [-6.79, -5.38]; P = <0.00001). The pooled studies were not heterogenous (I2 = 0%, P < 0.46). Conclusions: The lengthening of the humerus was regarded mostly as a cosmetic procedure. Using a variety of external fixators and intramedullary (IM) nails, the humerus was extended by distraction osteogenesis. There is a statistically significant link between length before and after humeral lengthening, although neither range of motion nor functional score are affected.

Keywords: Humeral; Lengthening; Fixators and Intramedullary.

1. Introduction

The functional and aesthetic effects of humeral deformity and shortening demand surgical treatment. [1, 2] A rising number of papers in the English language support the usual practice of bone lengthening and deformity treatment of the lower limbs. In contrast, there are few articles on the lengthening of the upper extremities. Dick and Tietjen reported the first instance of humeral lengthening in 1978. [3]

Due to the fact that the upper extremities of humans are non-weight-bearing, minor to moderate differences in arm length do not result in major functional drawbacks. Only when there is a moderate to substantial difference in arm length do practical or aesthetic limitations become apparent. Humeral hypoplasia has several origins, including both congenital and acquired defects. Among the reasons of disruption in early development include multiple exostoses, osteomyelitis, trauma, unicameral bone cysts, surgery, and radiation. Extreme differences in arm length may be handled in several ways, including no therapy, surgical lengthening, and epiphysiodesis proximal to the contralateral humeral epiphysis. [2]

The lengthening of the humerus was mostly seen as a cosmetic procedure. Recent research suggests, however, that the goals of bilateral humeral lengthening in achondroplasia are not just aesthetic, but also to restore proportions between the upper and lower limbs, expand reach, and improve the ability to perform perineal personal hygiene. [2] External fixators, such as multiaxial, uniaxial, and circular devices, have been utilized for this purpose, although intramedullary lengthening devices have just lately been produced. [4]

The goal of this study was to review the results of humeral lengthening in a systematic manner and met analysis of the results.

2. Methods

During this systematic review and meta-analysis preparation, we adhered to the PRISMA statement criteria and followed the Cochrane handbook for systematic reviews of interventions. [5, 6]

Search strategy and study selection

We searched Google Scholar, Egyptian Bank Knowledge (EKB) (Scopus and Web of science), PubMed on 20-28 March 2022 and updated the search on 24 April 2022 using appropriate keywords. The following search strategy was utilized for searching different databases: “(humeral AND lengthening OR deformity OR Ilizarov)”.

Study selection and eligibility criteria

Studies with criteria certain criteria were included: **(1)** Randomized control trails (RCTs), **(2)** English literature, **(3)** Retrospective Cohort, **(4)** Meta-analysis and Systematic review and **(5)** Outcomes either clinical or radiological are acceptable.

Exclusion criteria

We excluded conference abstracts, correspondence, unpublished data, non-English-language research, in-vitro investigations, animal and cadaver studies, and studies with unclear results or methodology reporting. All published publications were searched for search data with no constraints. Two-part screening of titles and abstracts was followed by screening of full-text documents. The reference lists of the included papers were carefully examined to uncover any further relevant research that may have been missed in earlier rounds.

Quality assessment

To evaluate the quality of the included studies, Methodological Index for Nonrandomized Studies (MINORS) checklist was used.

Data Extraction

We collected information from tables, text, figures (using graph grabber version 2.0), and additional data. We emphasized the following data: (Gender, age, length achieved, movement pre and post humeral lengthening).

3. Results

1. Literature search results: Initial results from the search generated 2,573 articles from three databases: 1,740 from Google Scholar, 750 from EKB (web of science= 457, Elsevier= 287), and 83 from PubMed. Of the 2573 listed articles. 1144 papers were eliminated due to duplication, 1108 articles underwent title and abstract screening, and 159 articles did not meet the inclusion criteria. The remaining 162 papers were reviewed in their entirety. In all, nine papers were considered in this systematic review and meta-analysis. **Fig. (1).**

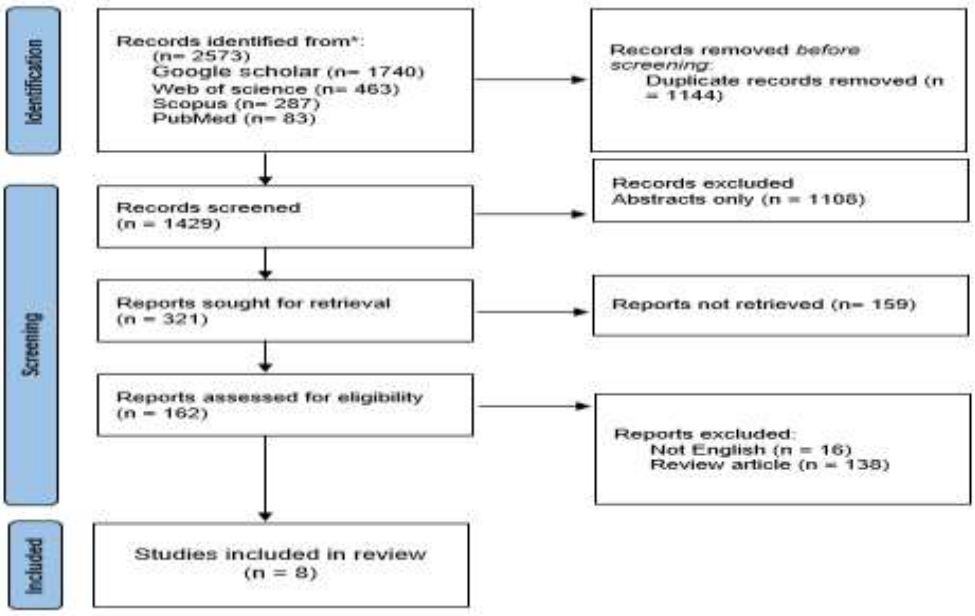

Fig. (1): PRISMA flow diagram of the literature search.

2. Quality assessment of the included studies: The Methodological Index for Nonrandomized Studies (MINORS) checklist was used to evaluate the quality of all included studies. The maximum MINORS score is 16 for studies. The MINORS score the 8 included studies ranging from 10 to 14 out of 16 **Table (4).**

3. Characteristics of the included studies: We identified eight studies discussing humeral lengthening including a total number of 158 humeri in 129 patients. The age range of patients across the studies ranged between 6.5 and 20 years. The male gender was common and ranging from 33% to 78%. Baseline characteristics of the included studies are described in **Table (1).**

4. Primary and secondary outcomes: Our primary outcome was to describe humeral lengthening including etiology, methods,

length achieved and effect on range of movement.

5. Outcomes: The humeral lengthening was conducted among 158 humeri among them 34 in the right side. The mean length achieved was 7.7 cm among the included eight145 studies ranging from 5.1 cm to 9 cm. **Table (2).**

Table (1): Basic characteristics of the included studies.

Study ID	Design	Sample size	Age, mean (SD)	Gender, n (%)
Hammouda [2]	Retrospective study	Six humeri and five patients	20 years	2 males (40%) 3 females (60%)
Hosny [3]	Retrospective study	56 segments in 46 patients	14 years	NM
Pawar [7]	Retrospective study	11 patients 15 humeral	24 years	five females (45%) and six males (55%)
McLawhorn [8]	Retrospective study	Three patients	10.3 ± 1.9 years	2 females (67%) 1 male (33%)
Kashiwagi [9]	Interventional study	20 humeri 10 patients	12 years	five females (50%) five males (50%)
Hosny [10])	Interventional study	16 patients	13 years (8.5–17 years)	nine girls (56%) and seven boys (44%)
Cattaneo [11]	Interventional study	43 humeral 29 patients	18 (10- 36 years)	16 male (55%) and 13 female (45%)
Stewart [12]	Rretrospective	13 humeri in 9 patients	13.4 (6.9 to 18.1) years	7 males (78%) 2 females (22%)

Table (2) : Side, Length achieved and range of motion among included studies.

Study ID	Side	Length achieved	Elbow movement pre	Elbow movement post
Hammouda	50% right, 50% left	5.1 cm	ROM 123	133
Hosny	NM	9 cm	DASH 15-40	7-16
Pawar	7 Right 8 left	7 cm	DASH 14.08	8.72
McLawhorn	2 left 1 right	6.5 ± 0.8 cm	NM	Improved in 2 patients
Kashiwagi	NM	7.8 cm (3.5 cm–10 cm)	75 to 110	80 to 110
Hosny	right sided in seven cases Left nine cases	8.6 (5.5 to 15 cm)	NM	Improved in 10 Maintained among 6 cases
Cattaneo	NM	9 (5- 16 cm)	NM	NM
Stewart	7 Right 2 Left	8.5 ± 1.3 cm EF 6.6 ± 2.3 cm in the MN	NM	NM

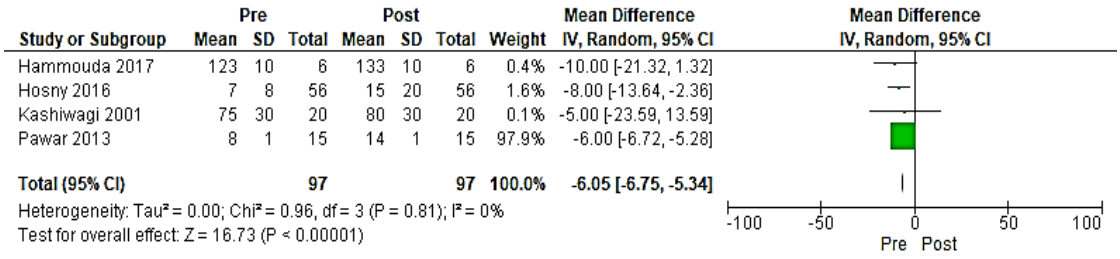

Fig.(2): Meta-analysis of pre and post humeral lengthening.

The pooled analysis of the included studies revealed a substantial correlation between pre- and post-lengthening humeral length. (OR = -6.08; 95% CI: [-6.79, -5.38]; *P* = <0.00001). The pooled studies were not heterogenous (I^2 = 0%, *P* < 0.46). Upon visual inspection of funnel plots, publication bias in these eight research was low (Figure 3). Using the standard error, the vertical axis of the graph estimated the sample size of the research. The horizontal spread indicated that the majority of studies were underpowered due to a broad CI of the effect size, which our research did not demonstrate.

Table 5 displays the results of the quality evaluation using the MINORS tool. The MINORS grade the eight included studies between 10 and 14 out of a possible 16.

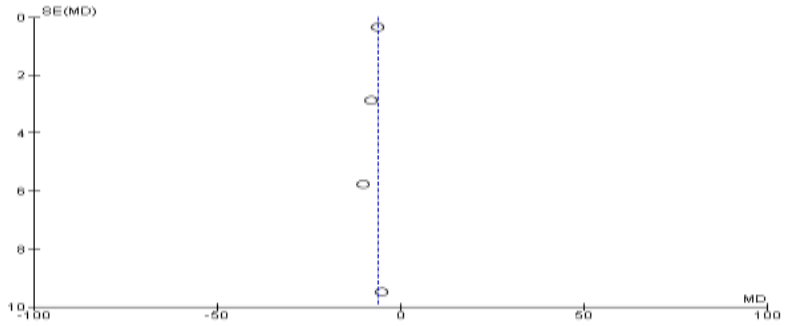

Fig.(3): Funnel plot for publication bias of the included studies.

Table (3): Etiology and follow up period of the included studies.

Study ID	Year	Etiology	Period	Follow up
Hammouda	2017	humeral growth arrest post-bone cyst postseptic growth arrest multiple hereditary exostosis	March 2014 to August 2015	1.8 years
Hosny	2016	(8 cases) epiphyseal injury, (10 patients) achondroplasia, (11 cases) infection and Erb's palsy (17 cases)	From 2002 until 2013	4.5 years
Pawar	2013	Achondroplasia Ollier's disease Unicameral bone cyst Childhood growth arrest Resection of malignant bone tumor	between 2001 and 2010	3.2 years
McLawhorn	2011	2 Osteomyelitis 1 Posttraumatic physeal arrest	-	2.5 years
Kashiwagi	2001	achondroplasia and only one case with hypochondroplasia	between 1996 and 1999	2.8 years (1.5–4.2 years)
Hosny	2005	Erb's palsy in 8 cases, epiphyseal injury in 5 cases and infection in three cases	From 1995 till 2001	3 years and 2 months (range 1 year and 4 months to 5 years and 6 months)
Cattaneo	1990	achondroplasia, old septic arthritis, birth palsy, fracture, congenital shortening, and benign neoplasm.	From 1982 to 1986	2.7 years (range, 0.5-5.5 years)

Table (3) Continue				
Stewart	2020	physeal arrest because of sepsis or trauma (9), achondroplasia (2), brachial plexus palsy (1), and unicameral bone cyst (1).	From 1999 to 2018	215 days

Table (4): Conclusion and quality of the included studies.

Study ID	Location	Conclusion	Method	Quality of study
Hammouda	USA	Effective and safe IM lengthening nails are available for humeral elongation. In particular, the PRECICE nail has fine control over the lengthening process.	IM PRECICE	Moderate
Hosny	Cairo	A legitimate treatment for deformity correction and arm shortening, including rotation and angulation, is humeral lengthening.	Ilizarov external fixation.	Moderate
Pawar	USA	Humeral lengthening with unilateral external fixation is an effective technique for improved patient function, with a complication rate equivalent to that of standard circular frames.	RAIL/ MAC	Moderate
McLawhorn	USA	The MAC system is appropriate for the correction of humeral length differences and associated humeral abnormalities in children.	Ilizarov	Moderate
Kashiwagi	Japan	For patients with achondroplasia, bilateral humeral lengthening was very helpful at enhancing function and proportion.	Ilizarov circular fixator	Low
Hosny	Egypt	The humerus is extinct. Without increasing the risk of shoulder instability, a single osteotomy can extend up to 115 percent of the original bone length.	Ilizarov	Moderate
Cattaneo	Canada	Using the Ilizarov technique, humeral lengthening may be safely done with great clinical outcomes.	Ilizarov technique	Moderate
Stewart	NM	The use of motorized intramedullary nails for humeral lengthening is a risk-free treatment that mitigates some side effects of EF, such as pin-site infection. It is well tolerated by patients. For substantial lengthenings, the reversal and reuse of MN may be considered.	Motorized nail	Moderate

Table (5) : Literature appraisal using MINORS assessment tool.

Study ID	A clearly stated aim	Inclusion of consecutive patients	Prospective collection of data	Appropriate endpoints	Unbiased assessment of study endpoint	Appropriate follow-up period	Loss to follow-up less than 5%	Prospective calculation of study size	Total
Hammouda	2	1	0	2	1	2	2	0	10
Hosny	2	2	0	2	2	2	2	0	12
Pawar	2	2	0	2	1	2	2	0	11
McLawhorn	2	2	0	2	1	2	2	0	11
Kashiwagi	2	2	0	2	1	2	2	0	11
Hosny	2	2	2	2	2	2	2	0	14
Cattaneo	2	2	2	2	2	2	2	0	14
Stewart	2	2	0	2	2	2	2	0	12

4. Discussion

The humerus lengthening was considered mostly a cosmetic treatment. [13-15] Recent study indicates, however, that the objectives of bilateral humeral lengthening in achondroplasia are not just cosmetic, but also to restore proportions between the upper and lower limbs, extend reach, and enhance the capacity to perform perineal personal hygiene. [16] External fixators, such as multiaxial, uniaxial, and circular devices, have been used for this purpose; however, intramedullary lengthening devices have been employed recently.

A single osteotomy level can yield 6 to 8 centimetres of regenerated bone, but successive osteotomies produce more length at the risk of developing soft tissue issues, such as joint stiffness and neuroprexia. If 10-16 cm of limb lengthening is required, it may be desirable to extend the child's limbs in two stages separated by 4 years.

This study aimed to conduct a systematic evaluation and meta-analysis of the findings of humeral lengthening studies published between 1990 and 2022.

Regarding the type of included studies, there were nine studies included. Six studies were retrospective, and three studies were intervention, with a total number of 158 humeri in 129 patients.

Major finding in the study were patient characteristic:

The age of patients across the studies ranges between 12 and 24 years. The lowest was found in the study of Kashiwagi [9]with range of 12 years and the highest was found in the study of Pawar [7] with range of 24 years. The male gender was common and ranging from 33% to 78%. There are no statically significant differences between the studies included regarding age and sex.

Follow up period

Follow up in the included studies range from 215 day in the study of Stewart in 2020 up to 4.5 year in the study of Hosny in 2016. [3] It was found that no results difference was noticed in short- and long-term follow-up.

Length achieved

The mean length achieved was 7.7 cm among the included nine studies ranging from 5.1 cm to 9 cm. The lowest was noticed in the study of Hammouda [2] with range about 5.1 cm using IM nail. The highest was noticed in the study of Hosny [3] in 2016 and the study of Cattaneo [11] with range about 9 cm using illizaroov external fixator. The pooled analysis of the included studies revealed a substantial correlation between pre- and post-lengthening humeral length. (OR = -6.08; 95% CI: [-6.79, -5.38]; P = <0.00001). The pooled studies were not heterogenous (I2 = 0%, P < 0.46). There is no statistically significant difference between the duration of the trials analyzed.

Range of movement (ROM)

ROM was only covered in three studies (Hammouda and Kashiwagi). [2, 9] There was no statistically significant difference between the included studies regarding pre- and post-operative range of motion, demonstrating that lengthening of the humerus had no effect on ROM regardless of the fixation technique used.

DASH score (functional score of shoulder)

The DASH questionnaire is a region-specific self-administered outcome tool designed to assess self-reported upper-extremity impairment and symptoms. The majority of the DASH consists of a 30-item disability/symptom scale, rated from 0 (no impairment) to 100. [17]

It was included only in three studies (Hosny in 2016 and Pawar). [3, 7] There is no statically significant difference between the

studies included regarding pre and post-operative functional score of shoulder which means that lengthening of humerus does not affect the functional score of shoulder.

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 [18].

On the other hand, it was also reported that retrograde approach could lead to elbow stiffness in fracture treatment [19].

Early physiotherapy during lengthening was suggested to prevent long-term limitation of the upper extremities [20].

The Ilizarov method and the monolateral fixators were shown to have a mean consolidation index of 27–32 days/cm [9, 20] and 24–32 days/cm, respectively [2].

In a systematic review by [18], the authors 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 [21].

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 [15, 22].

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 [10, 23]. Regarding the MIN, a mean humeral lengthening of 5.7 cm (5.0–7.5 cm) was obtained.

In [18] 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 [24].

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 [25]. This is similar to [18] 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, 21].

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 [18].

Other systematic reviews reported an infection rate of 0.8% with MIN lengthening in general (not specific to the humerus). This low rate of infection is due to the lack of communication between the lengthening device and the exterior environment [25].

Publication bias

Publication bias was minimal for those 8 studies included and most studies were underpowered.

5. Conclusion

The lengthening of the humerus was regarded mostly as a cosmetic procedure. Using a variety of external fixators and intramedullary (IM) nails, the humerus was extended by distraction osteogenesis. There is a statistically significant link between length before and after humeral lengthening, although neither range of motion nor functional score are affected.

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