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Pediatric and Juvenile Lunatomalacia: To Treat Surgically or Not? A Systematic Review and Personal Experience

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Background: Pediatric or juvenile lunatomalacia is still a less understood disease of childhood or young adolescence with spontaneous lunate osseous alterations and sometimes incomplete reconstitution of the lunate shape and bony structure. The treatment regimen to be used is still unclear. Given the young age of the patients, the question arises whether surgery is beneficial.
Methods: We performed a systematic literature review and analysis. We included studies from the last 5 decades addressing this issue and included additional publications identified through manual screening of references. Data were collected from public bibliographic databases.
Results: We included a total of 37 studies in this article. No studies of higher levels of evidence were available. Almost all were case reports or case series, but the quality was overall rated good in most cases. Adding our case to the available studies with a focus on the treatment results, the survey and statistical analysis revealed that a conservative approach most likely results in the best outcomes until the age of 15 years.
Conclusions: Our systematic review of this topic demonstrated a lack of high-quality studies. Only reports and smaller case series were found to provide answers. The total number of patients with this entity is also low, but this work is the most comprehensive review of reported cases in the literature. Considering all these reports and our case, we recommend a conservative approach until the age of 15 years.
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INTRODUCTION

Pediatric or juvenile lunatomalacia is a poorly understood disease of the lunate with fracturing, collapse, reconstitution, and often complete remission at a younger age, in contrast to the clinical course of avascular lunate necrosis in adults, known as the Kienböck disease. The cause is speculative both in adults and even more so in pediatric or adolescent patients. All existing theories have never been validated. It can, therefore, be assumed that pediatric lunatomalacia has a different clinical course than lunate necrosis in adults. Treatment should therefore also be different. In fact, there is still uncertainty about which type of treatment—such as radial wrist shortening—is most appropriate at which age in pediatric and adolescent patients, and when to move from one treatment to another. We ourselves had a case of juvenile lunatomalacia at our own institution that raised the question of the right treatment. There is no high-quality literature to help clinicians make decisions based on patient demographics and long-term treatment outcomes. For this reason, we undertook a systematic review of all known published cases of pediatric or juvenile lunatomalacia to attempt to address this question.

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METHODS

This article adheres to the preferred reporting items for systematic reviews and meta-analyses 2020 guidelines[1] with secondary exceptions in the Discussion section. The Prospero database was checked before the search for existing reviews and has been registered there.

Search Strategy and Selection Criteria

A systematic search through 3 different public databases, PubMed, Scopus, and Web of Science, was conducted. The first data were extracted by search queries in February 2023: "juvenile" AND "lunatomalacia" OR

Disclosure statements are at the end of this article, following the correspondence information.

"pediatric" and "lunatomalacia" OR "juvenile" AND "Kienbock" OR " pediatric" AND "Kienbock." All terms were identically used in all 3 databases. All studies found were screened for duplicates, and these were removed before further processing. Additional publications found by manually screening references and citations of studies intended for inclusion were also used in this publication. An updated search was performed in January 2024. Studies downloaded from the 3 databases were assessed for quality and further selection. No language restrictions were included in the initial search. Data extraction was guided by the previously prepared protocol and conducted by the authors (see following sections). Case reports or case series with a low number of patients were also included. No study was excluded based on the initial search by quality. A search for unpublished work was not performed. Two independent reviewers screened titles and abstracts to be sufficient for inclusion in this article. Disagreement was resolved by consensus with full-text screening. Studies only available as abstracts were not included in this review. Additional studies were also reviewed in the same way by the 2 independent reviewers. Studies with languages other than English were excluded. They were translated into English by Google Translate and then discussed in the Discussion section at the end of this article.

Quality of Studies

Two independent reviewers assessed the quality of the included studies, and every study was rated as poor, fair, good, or not applicable. Disagreement was solved by consensus via internal discussion. The quality was assessed by the Joanna Briggs Institute (JBI, Adelaide, South Australia) critical appraisal tool for case series studies, the JBI critical appraisal checklist for case reports, and the bias by the risk of bias in systematic reviews (ROBIS) tool, following the summary of phase 2 ROBIS domains, phase 3, and signaling questions[2] and checklist.

Data Extraction

The extraction protocol was based on data estimated as relevant to this article. We estimated the title of the study, abstract, full-text availability, diagnosis, date of publication, number of patients included, patient age at the time of treatment, dominance, affected side, sex, type of treatment, complications, duration of immobilization, Lichtman stage, follow-up time, ulnar variance before treatment, epiphyseal state, study type, type of intervention, reported result of the treatment, and possible cause of the disease. All data were manually extracted by the authors.

Data Analyses

For each included study, we collected all available data given by the different authors of the publication. Existing data and text information were collected in Microsoft Excel, and results were presented in tabular form; statistical analyses of a possible cut-off point of conservative treatment related to age and visualization were evaluated by the software package R. The R code for analysis was programmed by the first 2 authors. Methodological statistical analyses were previously discussed with a biostatistician.

Takeaways

Question: What is the appropriate treatment of pediatric or juvenile lunatomalacia depending on the patient's age?

Findings: With a systematic review and our own personal experience, we found a conservative approach most appropriate for those younger than the age of 15 years.

Meaning: Treat pediatric and juvenile lunatomalacia patients younger than the age of 15 years conservatively with splinting up to 4 months depending on the clinical presentation.

RESULTS

Inclusion of Studies

With this search strategy, we found 18 publications on PubMed from 1999, 120 on Scopus from 1975, and 17 on Web of Science from 2003, totaling 155 publications. After removing duplicates and screening titles for subject relevance, 25 studies remained. Three of these 25 were removed for not fitting the inclusion criteria; thus, 22 studies remained for further processing. By screening the references of these 22 studies, we found an additional 22 studies not identified in the public database search. Of these studies, 6 were written in a language other than English, 1 showed insufficient data, and 1 was only available as an abstract. In summary, 37 publications could be included in our review.

Study Characteristics

Study characteristics are summarized in Table 1. The studies, we included, were published between 1984 and 2023 (see Fig. 1). None of the studies had a high level of evidence, for example, a randomized double-blinded control trial. All of the publications were case series or even case reports without controls. Inclusion criteria were the presence of lunatomalacia and patients 18 years of age or younger.

Data Analysis

For each included study, we noted the authors, date of publication, patient age, dominance, side of wrist pain, sex, treatment, complications, immobilization period, and Lichtman stage. Data were collected in Microsoft Excel, and the results were presented in tabular form. Excluded studies were separately reviewed in the Discussion section. Data and statistical analyses were performed using the software package R ×64 v4.1.3.[40] The single case in the database with bilateral lunatomalacia was counted for statistical analysis as 2 independent cases.[35]

Quality Assessment

Quality assessment of the case series was based on the JBI critical appraisal tool for case series[41] and the JBI checklist for case reports (see Table 1). Case series (n = 6), which were based on complete data were rated good (n = 4), and case series based on missing data were rated poor (n = 1) or rated fair (n = 1). Furthermore, most case series reported a limited number of patients ranging from

Table 1. List of All Selected English Publications

Author	Year of Publications	Patient Number	Age of the Patient, y	Dominance	Affected Side	Sex	Immobilization (Weeks) Without Operation or Until Operation	Lichtman Type	Ulnar Variance Pretreatment, mm	JBI Quality Tool Result for Case Reports and Series
Lanzer et al[3]	1984	1	18	Right	Right	Male		Lichtman III B		Fair
Rasmussen et al[4]	1986	2	8		Right	Female	56	See supl.	See supl.	Good
Amadio et al[5]	1987	3	14	Right	Left	Male		Lichtman III	0.0	Good
Edelson et al[6]	1988	4	12	Right	Right	Female	6	See supl.	−5.0	Good
Hosking[7]	1989	5	8		N/a	Male	24	Lichtman III	−5.0	Good
Nakamura[8]	1990	6	16		N/a	Male		Stahl stage II	0.0	Good
Nakamura[8]	1990	7	17		N/a	Male		Stahl stage IV	2.0	Good
Nakamura[8]	1990	8	10		N/a	Male		Stahl stage IV	−3.0	Good
Nakamura[8]	1990	9	18		N/a	Male		Stahl stage II	−2.0	Good
Nakamura[8]	1990	10	16		N/a	Female		Stahl stage IV	−2.5	Good
Nakamura[9]	1991	11	16		Right	Male		Lichtman II	0.0	Good
Nakamura[9]	1991	12	15		Left	Female		Lichtman IV	−2.5	Good
Nakamura[9]	1991	13	17		Right	Male		Lichtman II	−2.0	Good
Nakamura[9]	1991	14	9		Right	Male		Lichtman IV	−3.0	Good
Nakamura[9]	1991	15	13		Right	Female		Lichtman IV	0.0	Good
Nakamura[9]	1991	16	16		Left	Male		Lichtman IV	0.0	Good
Nakamura[9]	1991	17	17		Right	Male		Lichtman II	1.0	Good
Imaeda et al[10]	1992	18	15		Right	Female		Lichtman III		Fair
Kahn and Sherry[11]	1994	19	7		Left	Female		See supl.		Fair
Foster[12]	1996	20	8	Right	Left	Female		Lichtman III	See supl.	Good
Greene[13]	1996	21	11		Left	Female	26	See supl.		Good
Yasuda et al[14]	1998	22	12	Right	Left	Female	8	Lichtman IIIB	−0.5	Good
de Smet[15]	2003	23	12	Right	Right	Female		Lichtman III	−3.5	Good
Ferlic et al[16]	2003	24	13		Left	Male		Lichtman IIIA	−3.0	Good
Cvitanich and Solomons[17]	2004	25	8		Left	Male	18	Lichtman IIIB	See supl.	Good
Schweizer et al[18]	2004	26	13		Left	Female	4	Lichtman II	−2.5	Good
Shigematsu et al[19]	2005	27	11	Right	Right	Female	12	Lichtman IIIA		Good
Herdem et al[20]	2006	28	15		Right	Male	24	See supl.	−2.0	Good
Herzberg et al[21]	2006	29	14	Right	Right	Female	12	See supl.	0.0	Good
Kazuki et al[22]	2006	30	15	Right	Right	Female		Lichtman IIIA		Good
Hurley and McKee[23]	2008	31	13	Right	Right	Female		Lichtman IIIA	−3.0	Good
Ando et al[24]	2009	32	12	Right	Left	Female	8	Lichtman IIIB	−0.5	Good
Ando et al[24]	2009	33	14	Right	Left	Female	8	Lichtman IIIA	−1.0	Good
Ando et al[24]	2009	34	15	Right	Right	Female	32	Lichtman IIIA	0.0	Good
Ando et al[24]	2009	35	16	Right	Left	Male		Lichtman IIIA	0.0	Good
Ando et al[24]	2009	36	9	Right	Right	Female		Lichtman IIIB	−2.0	Good
Ando et al[24]	2009	37	17	Right	Left	Male		Lichtman IIIB	1.0	Good
Docquier et al[25]	2009	38	6	Right	Left	Male	22	Lichtman IIIA	−4.0	Good
Matsuhashi et al[26]	2009	39	12		See supl.			Lichtman II	See supl.	Fair
Matsuhashi et al[26]	2009	40	18		See supl.			Lichtman II	See supl.	Fair
Matsuhashi et al[26]	2009	41	11		See supl.			Lichtman IIIA	See supl.	Fair
Matsuhashi et al[26]	2009	42	13		See supl.			Lichtman IIIA	See supl.	Fair
Matsuhashi et al[26]	2009	43	12		See supl.			Lichtman IIIB	See supl.	Fair
Matsuhashi et al[26]	2009	44	13		See supl.			Lichtman IIIB	See supl.	Fair
Matsuhashi et al[26]	2009	45	17		See supl.			Lichtman IIIB	See supl.	Fair
Matsuhashi et al[26]	2009	46	18		See supl.			Lichtman IIIB	See supl.	Fair
Irisarri et al[27]	2010	47	9			Female				Poor
Irisarri et al[27]	2010	48	11			Male				Poor
Irisarri et al[27]	2010	49	11			Male				Poor
Irisarri et al[27]	2010	50	12			Female				Poor
Irisarri et al[27]	2010	51	13			Female				Poor
Irisarri et al[27]	2010	52	14			Male				Poor
Irisarri et al[27]	2010	53	14			Male				Poor
Irisarri et al[27]	2010	54	15			Female				Poor

(*Continued*)

Table 1. *Continued*

Author	Year of Publications	Patient Number	Age of the Patient, y	Dominance	Affected Side	Sex	Immobilization (Weeks) Without Operation or Until Operation	Lichtman Type	Ulnar Variance Pretreatment, mm	JBI Quality Tool Result for Case Reports and Series
Irisarri et al[27]	2010	55	15			Female				Poor
Irisarri et al[27]	2010	56	16			Female				Poor
Irisarri et al[27]	2010	57	16			Male				Poor
Irisarri et al[27]	2010	58	16			Male				Poor
Irisarri et al[27]	2010	59	16			Male				Poor
Shayesteh Azar et al[28]	2011	60	11	Right	Right	Female	6	See supl.		Good
Jorge-Mora et al[29]	2012	61	13		Right	Male		Lichtman IIIB	−10.0	Good
Jorge-Mora et al[29]	2012	62	14		Bilateral	Male		Lichtman I	−1.0	Good
Jorge-Mora et al[29]	2012	63	12		Left	Female		Lichtman I	0.0	Good
Jorge-Mora et al[29]	2012	64	12		Left	Female		Lichtman IIIA	−5.0	Good
Shimizu et al[30]	2013	65	14		Bilateral	Male	8	Lichtman IIIA	See supl.	Good
Farr et al[31]	2014	66	15			Female		Lichtman IIIB	See supl.	Good
Farr et al[31]	2014	67	15			Male		Lichtman IIIB		Good
Omor et al[32]	2015	68	17	Left	Left	Male		Lichtman III		Fair
Campbell et al[33]	2017	69	17		Left	Female	8	Lichtman I		Good
Afshar[34]	2018	70	15		Right	Male	6	Lichtman III		Good
Afshar[34]	2018	71	13		Right	Female	6	Lichtman III		Good
Huang et al[35]	2019	72	14	Right	Bilateral	Male		See supl.	See supl.	Good
Mohseni et al[36]	2019	73	11	Right	Left	Female	10	Lichtman IIIA		Good
Lichtman et al[37]	2022	74	16				12	Lichtman I	See supl.	Poor
Al-Zoubi et al[38]	2023	75	11		Left	Male		See supl.		Good
Lendrum et al[39]	2023	76	7	Right	Right	Female	12	See supl.		Good

Reduced table content of non-English published case reports in the literature with our own case.

4 to 13 individuals. Case reports (n = 31) were most often rated as good (n = 26), with only 4 rated fair and 1 poor. Statistical power analysis was impossible due to the lack of randomized controlled trials (RCTs) and inconsistent or missing data.

Bias assessment was carried out using the ROBIS tool.[2] We estimated the risk of selection bias as high because of the low number of publications found in the most common public databases with our search query. Thus, the representativeness of the included article is low. Publication bias has been estimated as high because of the low number of cases reported worldwide. The studies themselves were heterogeneous, and therefore, the risk of bias in the synthesis of findings was rated high.

Studies

Our search strategy revealed a total of 177 articles. Manually found articles by screening references were added, but only 37 were eligible for further processing. Study characteristics are summarized in Table 1. None was a randomized double-blinded control trial. The remaining publications were all case series or case reports and, overall, of different quality. Statistical analysis was performed, but no meta-analysis was conducted due to the absence of RCTs. Due to the very low prevalence, an RCT for this disease is impossible to perform. An international cohort study or a case-control study would probably be a valid option to find out whether conservative treatment is the better alternative and up to what age. The most important thing is to create a complete data set for each patient treated. The inclusion criteria of patients enrolled in case reports/case series was a radiological diagnosis of lunatomalacia. The studies we included were published between 1984 and 2023. Screening our own single-institution patient database, we found only 1 patient with this unusual disease over the last decade. Altogether, we found 86 patients with pediatric/juvenile/adolescent lunatomalacia. Of those, we excluded 10 and proceeded with 76 for further statistical processing. Of the 76 patients, only 1 had a consecutive lunatomalacia bilaterally.

Data Analysis

Visualization and statistical data analyses were done with the software package R. The age of the included patients ranged from 6 up to 18 years, with a mean of 13.4 years (Fig. 2). One patient was left-handed (1%), 22 were right-handed (29%), and the rest of the data were missing. Of all patients, 21 had the affected hand on the left side, 22 on the right, 32 not stated, and 1 patient was affected bilaterally. Of the 76 patients, 34 were men (45%), 33 were women (43%), and in 9 patients, their sex was not mentioned. The ulnar variance ranged between −10 and 2 mm with a mean of −1.9 mm (Fig. 3).

Twenty-eight hands were treated conservatively, 31 operatively, and 18 combined (splinting followed by operation). The patient affected bilaterally received an identical operative procedure on both sides (Fig. 4). Of those who received a period of immobilization, the duration ranged from 4 weeks up to 56 weeks, with a mean of 14.9

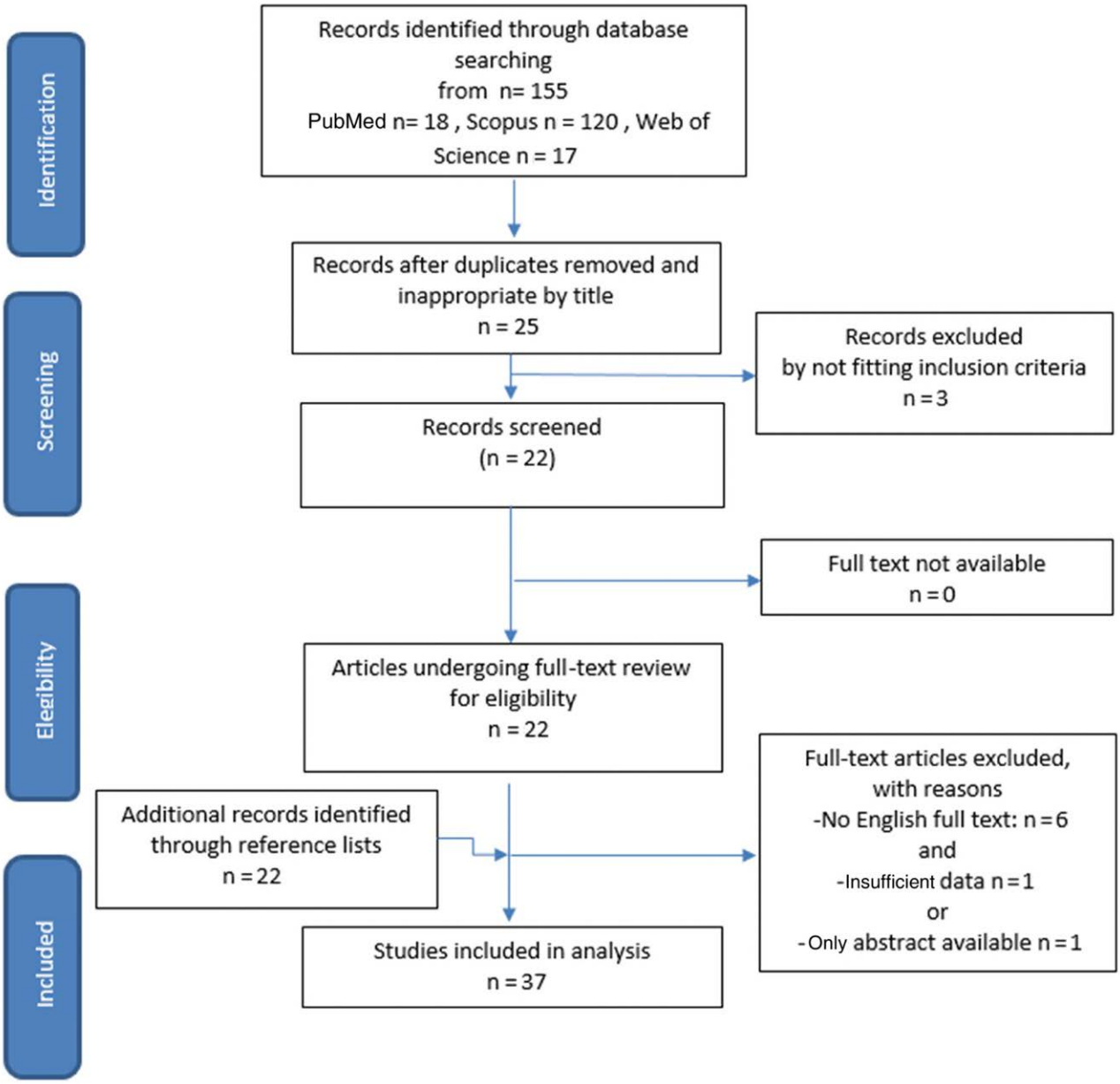

Fig. 1. Flow diagram of the study selection process.

weeks (Fig. 5). In 55 patients, this information was missing. The total follow-up time ranged between 4 and 180 weeks with a mean of 38.5 weeks. For 15 cases, the follow-up time was missing. Of the 77 treated hands, 9 had a reported complication (Fig. 6).

Of the patients who were treated conservatively, the mean age was lower (mean 12.5 y) compared with those who directly received an operative treatment (mean 15 y) (Fig. 7). Figure 8 demonstrates the probability of developing a complication is higher in younger patients. When comparing the ages of those who underwent surgery and those who did not, an analysis of variance test followed by a paired t test with Bonferroni P adjustment for multiple comparison correction yielded a P value of 0.021 ($P < 0.05$ for statistically significant) for complications in the surgical treatment group with younger age. This means that surgery in young children has a significantly higher rate of complications than in older patients or those who did not undergo surgery. Testing the association between complications and surgery using the Pearson chi-square test returned a P value of 0.052. The Pearson chi-square test for testing the association between Lichtman type and complications resulted in a P value of 0.424 ($P < 0.05$). It means there is no statistical correlation between the Lichtman stage and complications. Further statistical analyses were not possible with the incomplete data set available.

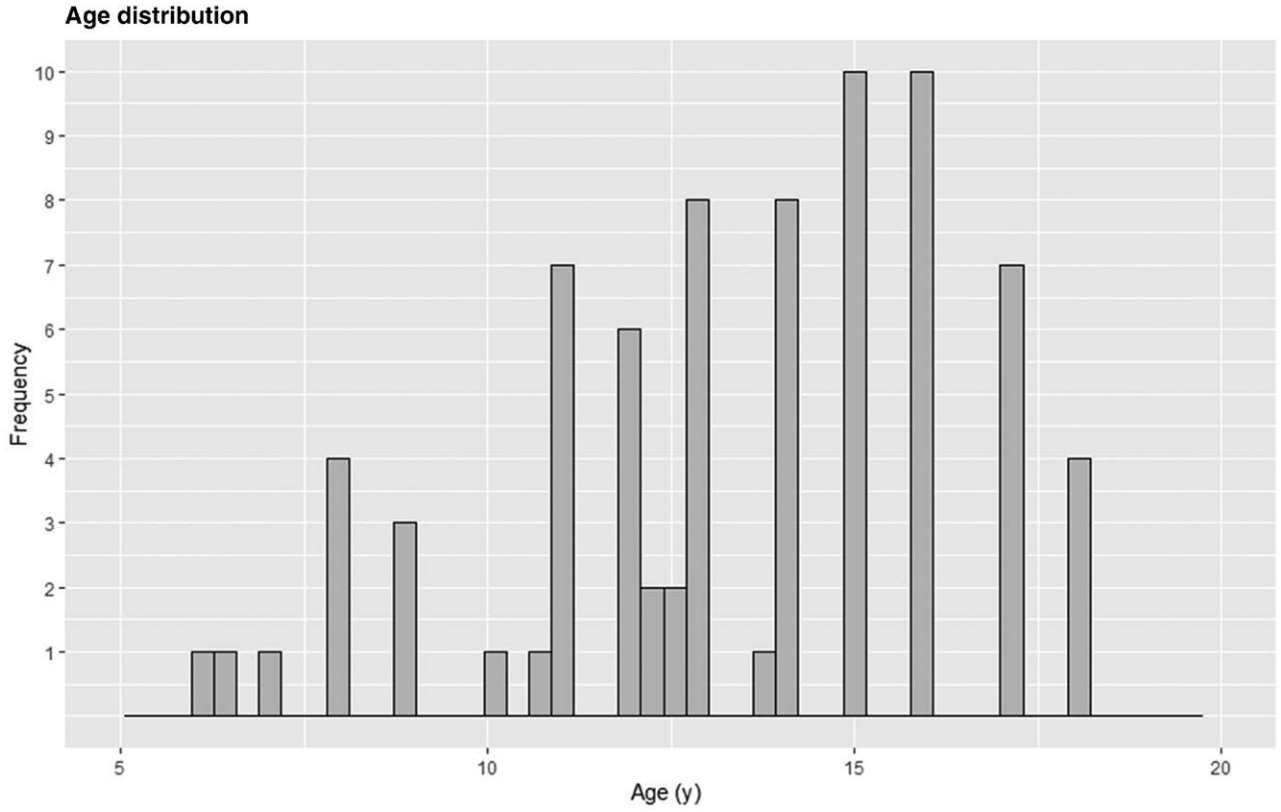

Fig. 2. Age distribution of the published patients.

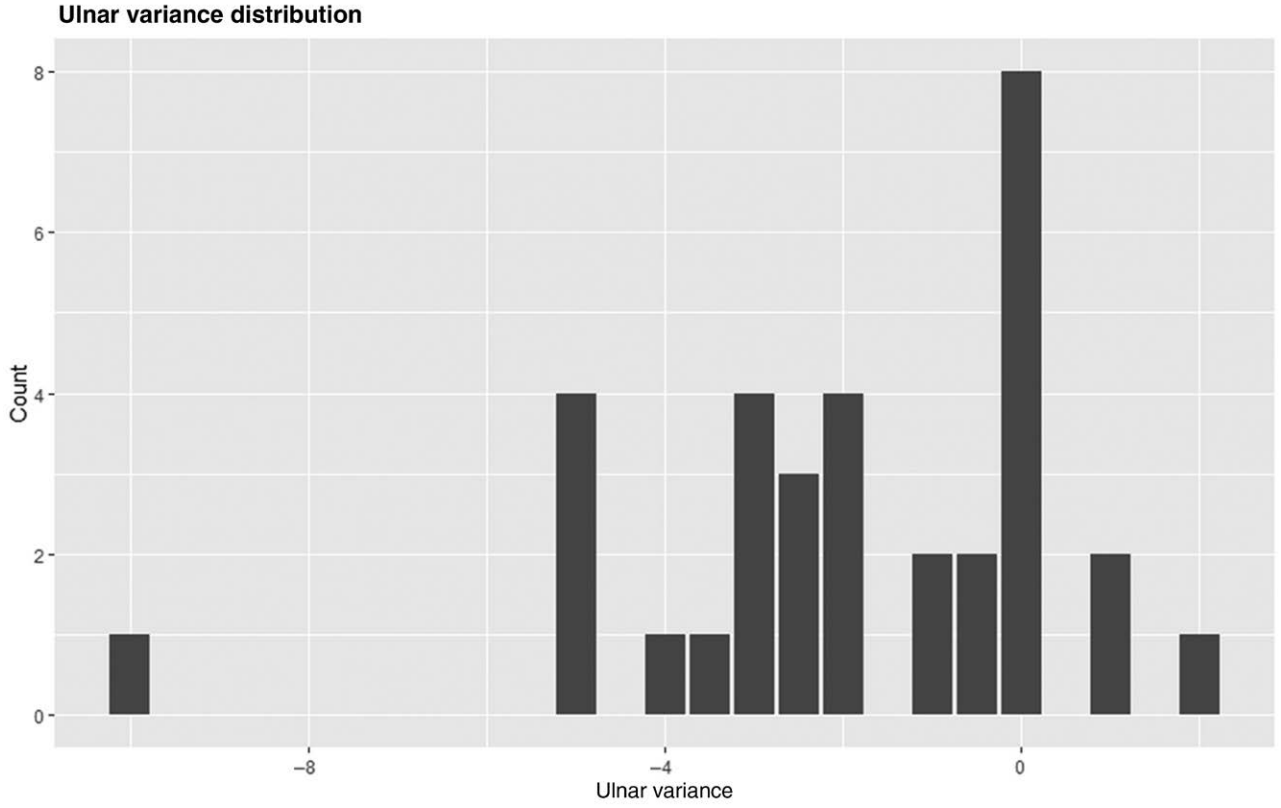

Fig. 3. Ulnar variance distribution.

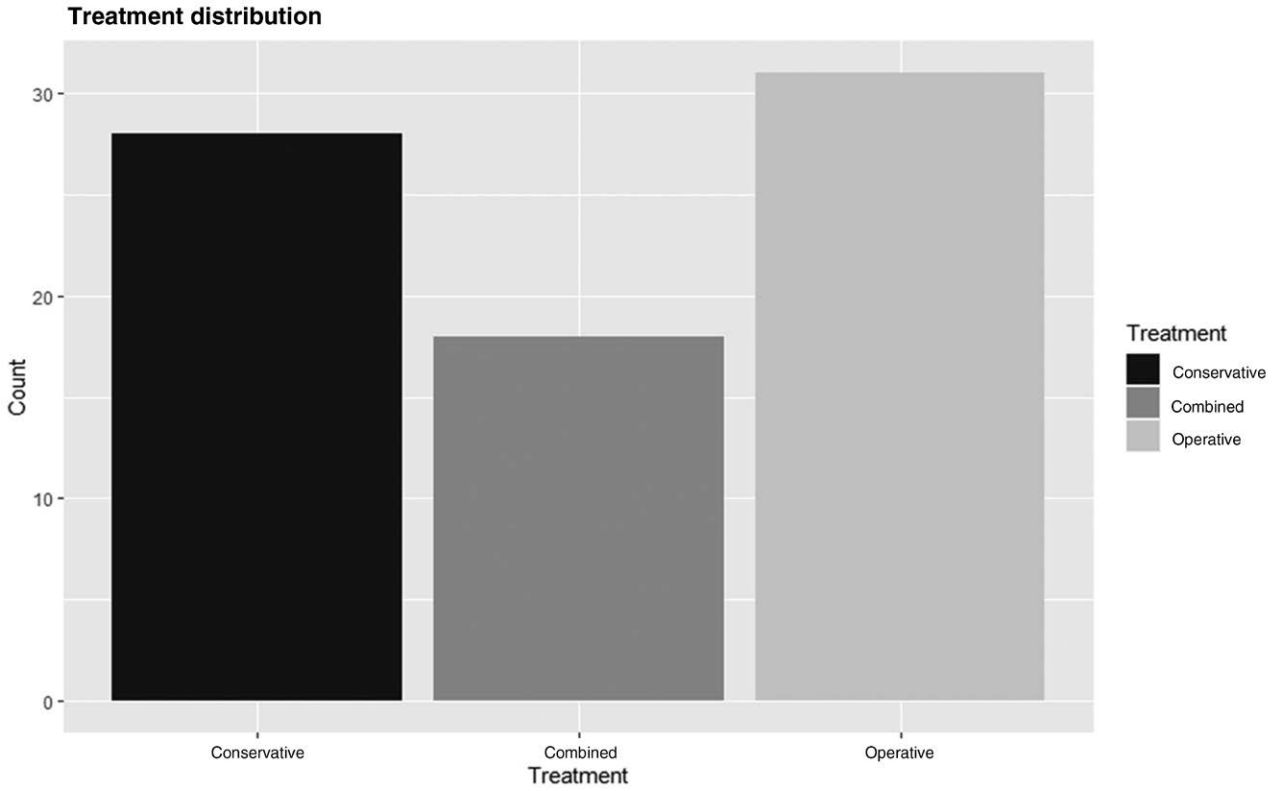

Fig. 4. Conservative, operative, and combined treatment distribution.

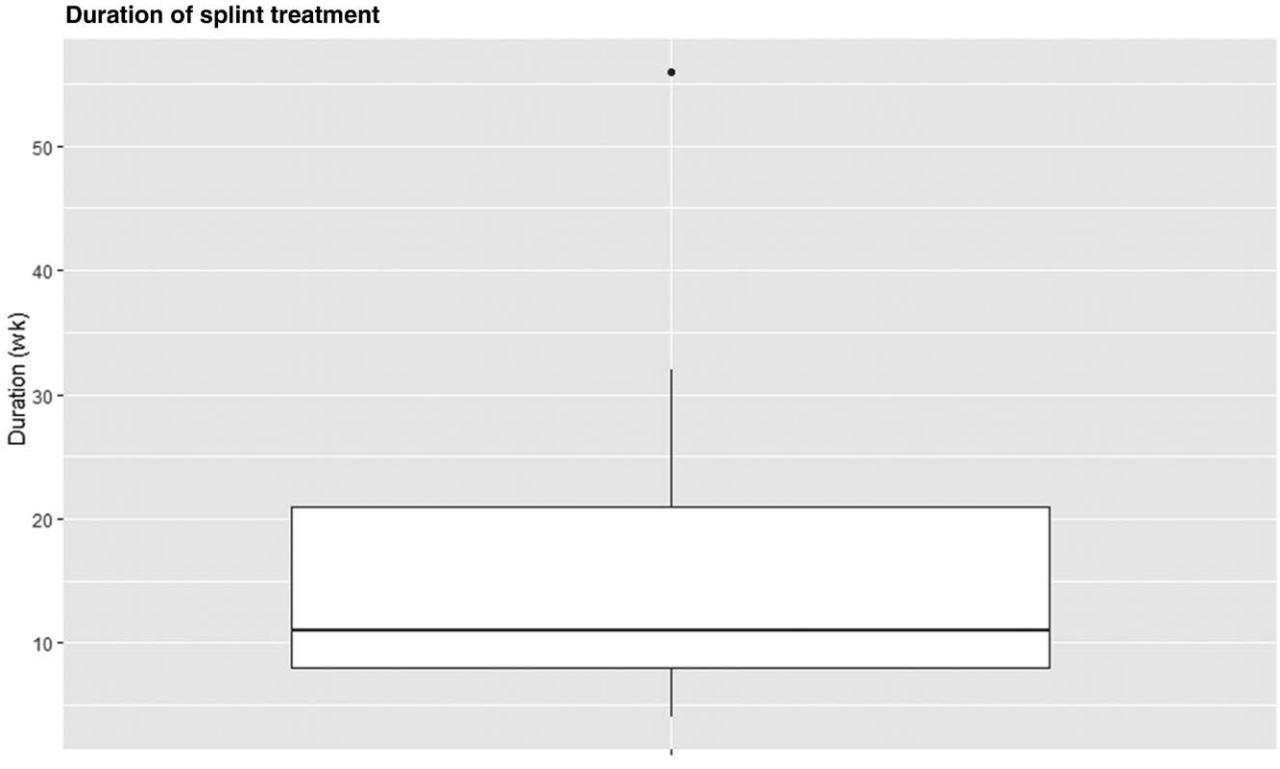

Fig. 5. Duration of splint treatment in weeks.

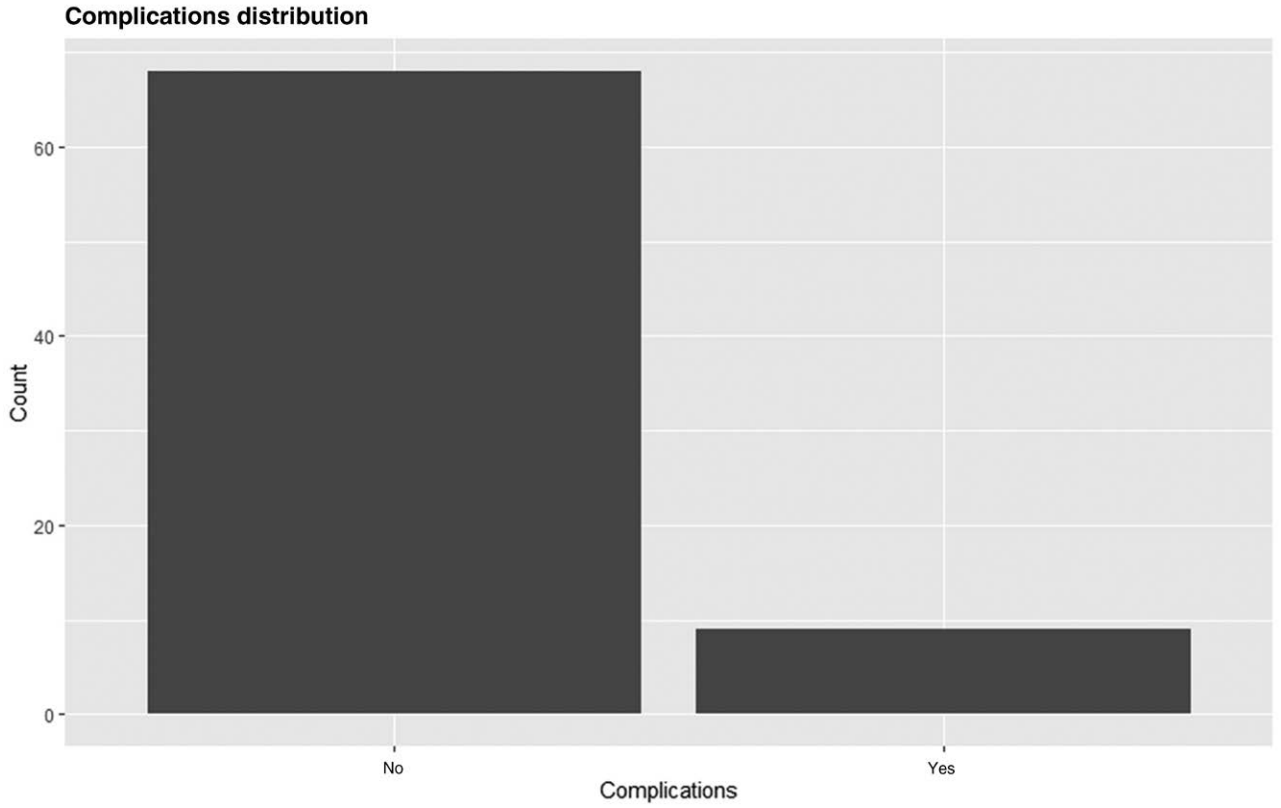

Fig. 6. Distribution of complications.

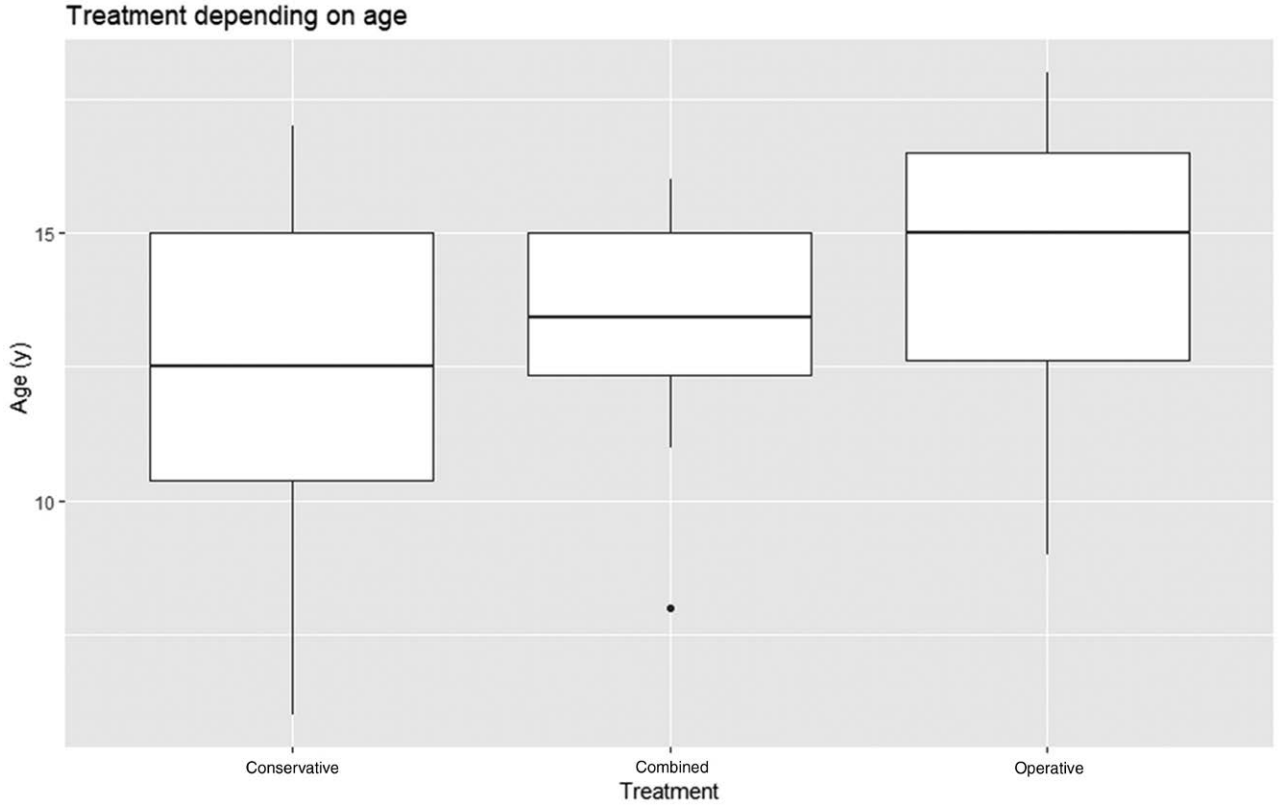

Fig. 7. Distribution of patient age and treatment.

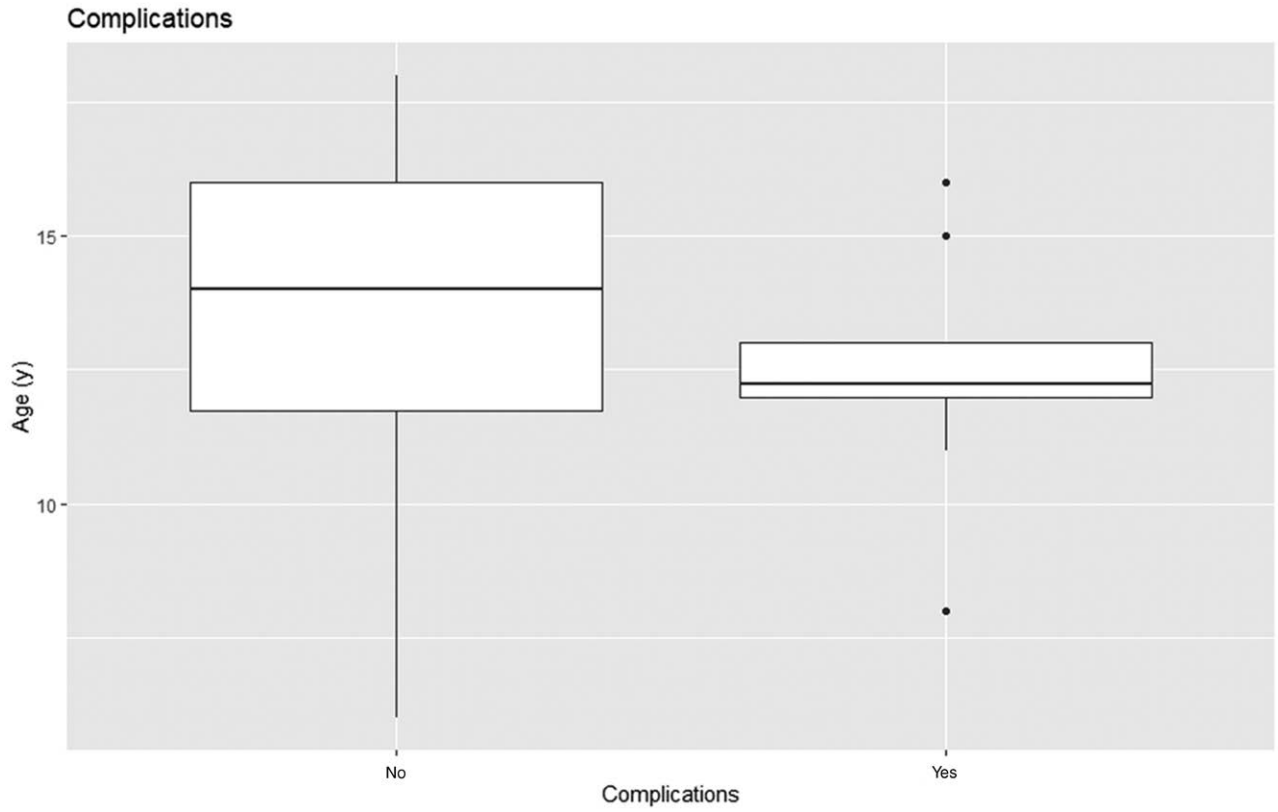

Fig. 8. Complication in relation to patient age.

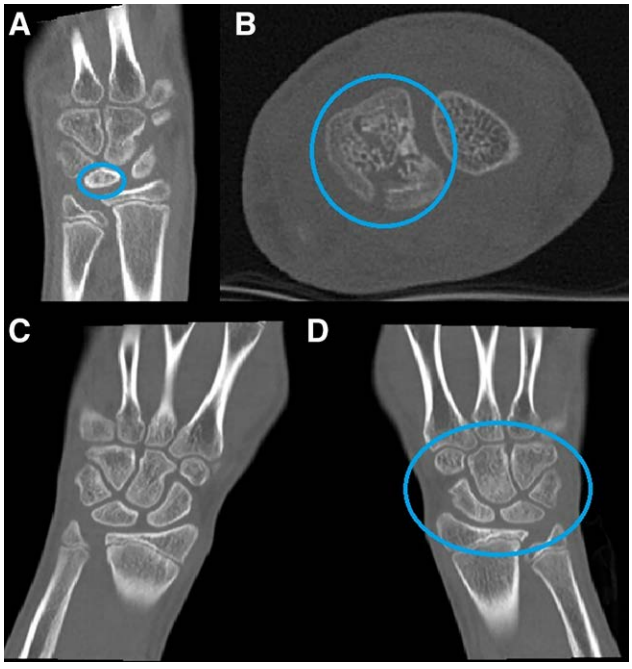

Fig. 9. Control CT scans of our patient during the clinical course. CT scan of the left wrist demonstrates Lichtman stage II at first presentation (A) (blue circle), a coronary CT scan at follow-up 4 months later demonstrates fragmentation according to stage Lichtman IIIA (B), and complete reconstruction of the lunate at 12 months after first presentation (C and D. The left wrist is on the right side of the scan visible).

Individual Studies

Lanzer et al[3] first reported juvenile lunatomalacia in 1984 in a patient younger than 19 years with sickle cell anemia, treated surgically with proximal row carpectomy after progression to Lichtman stage III. Rasmussen et al[4] described the first case of a child with complete resolution after conservative treatment. Amadio et al[5] presented a case of a 14-year-old boy who was treated conservatively initially, and subsequently with a Herbert screw and an external fixator. Edelson et al[6] reported a recurrence in a 12-year-old girl after previous treatment with radial shortening. Hosking[7] published a case of an 8-year-old boy treated only conservatively. The first case series was presented by Nakamura et al[8,9] in 1990; 5 patients, 10–18 years of age with Stahl classification between II and IV, were all directly operated on with an excellent result. In 1991, Nakamura published a case series of young, physically active patients who received different types of treatment from conservatively by Lichtman stage IV to proximal row carpectomy by Lichtman stage II disease. Imaeda et al[10] described a 15-year-old girl (stage III) who underwent radial osteotomy. In 1994, Kahn and Sherry[11] reported a case of a 7-year-old girl with lunatomalacia Lichtman stage III after corticosteroid treatment for dermatomyositis. Foster[12] documented an 8-year-old girl treated conservatively before undergoing radial shortening with postoperative complications. Greene[13] demonstrated complete resolution in an 11-year-old with cerebral palsy via conservative therapy. Yasuda et al[14] described a 12-year-old girl

Table 2. Reduced Table Content of Non-English Published Case Reports in the Literature With Our Own Case

Author	Year of Publication	Patient Number	Age of the Patient, y	Dominance	Affected Side	Sex	T (0 = Conservative, 1 = Conservative + Operative, 2 = Operative)	Complications of Treatment	Immobilization (Weeks) Without Operation or Until Operation	Lichtman Type	Follow-up	Ulnar Variance Pretreatment	Epiphyseal Plate	Type of Study	JBI Quality Tool Result for Case Reports and Series
Benz and Blencke[45]	1976	77	7	N/A	N/A	Female	0		9	N/A	26	N/A	N/A	Case report	Poor
Da Costa[46]	1993	78	10	N/A	Right	Male	2		N/A	N/A, probably Lichtman II	20	N/A	Open	Case report	Fair
Queiroz et al[47]	1997	79	11.16	N/A	N/A	Female	1		4	Lichtman IIIA	24	0 mm	N/A	Case report	Good
Queiroz et al[47]	1997	80	14.42	N/A	Right	Female	1		4	Lichtman IIIA	15	−0.5 mm	N/A	Case report	Good
Mathieu and Dumontier[48]	2009	81	9	Right	Left	Male	0		24	Lichtman II	15	−3 mm	Open	Case report	Good
Kalb et al[49]	2010	82	12	N/A	Left	Female	1		N/A	Lichtman IIIA	102	N/A	N/A	Case report	Poor
Kalb et al[49]	2010	83	11	N/A	Left	Female	0		24	Lichtman I	24	N/A	N/A	Case report	Poor
Kalb et al[49]	2010	84	12	N/A	Left	Female	1	K-wire dislocation	N/A	Lichtman IIIB	18	N/A	N/A	Case report	Poor
Kim et al[50]	2018	85	14	N/A	Left	Male	0		12	Lichtman IIIB	24	0 mm	Open	Case report	Good
Current study	2023	86	12	Right	Left	Male	**0**		**30**	**Lichtman II->IIIA**	**12**	**−4 mm**	**Open**	**Case report**	**Good**

(stage IIIB) successfully treated with temporary K-wire fixation. De Smet[15] reported of a 12-year-old girl treated conservatively. In 2003, Ferlic et al[16] reported a case of a 13-year-old boy with Lichtman stage IIIA, which was treated with radial shortening osteotomy. Cvitanich and Solomons[17] reported an 8-year-old boy who fully recovered after conservative management. In 2004, Schweizer et al[18] reported a 13-year-old girl (stage II) treated with immobilization and calcitonin therapy. Shigematsu et al[19] described an 11-year-old girl (stage IIIA) treated with temporary scaphotrapezoidal joint fixation, resulting in revascularization and symptom resolution. Herdem et al[20] documented a 15-year-old boy (stage IIIB) initially treated conservatively but requiring vascularized bone grafting and radial shortening due to disease progression. Herzberg et al[21] and Kazuki et al[22] described successful outcomes with conservative treatment and K-wire fixation, respectively, in adolescents. Hurley and McKee[23] presented a 13-year-old girl (stage IIIA) treated with radial shortening. Ando et al[24] described a case series of 6 adolescents treated with K-wire fixation, with excellent outcomes but some complications. Docquier et al[25] reported the youngest case, a 6-year-old treated with an orthosis, resulting in complete lunar reconstruction. Matsuhashi et al[26] presented 8 cases treated with radial shortening, 4 of which developed reactive radial overgrowth. Irisarri et al[27] reported 13 cases, with conservative treatment for most and surgery (radial shortening) in 3 older patients. Shayesteh Azar et al[28] documented an 11-year-old (stage IIA) with full resolution after conservative treatment. Jorge-Mora et al[29] introduced distal radius epiphysiodesis for 4 patients, achieving symptom-free outcomes. In 2013, Shimizu et al[30] reported bilateral lunatomalacia in a 14-year-old boy, managed conservatively on 1 side and surgically (K-wire fixation) on the other. Farr et al[31] described vascularized bone graft transfer and external fixation in 2 cases with good results. Omor et al[32] documented a 17-year-old (stage III) treated conservatively with unknown outcomes. In 2017, Campbell et al[33] reported conservative treatment of a 13-year-old girl, likely stage I, with symptom improvement. Afshar[34] described 2 adolescent athletes treated conservatively, 1 requiring surgery due to persistent pain. Huang et al[35] reported a 14-year-old boy with bilateral disease (stages II and IIIA) successfully treated with grafting. Mohseni et al[36] described an 11-year-old girl (stage IIIA) with resolution after splinting. Al-Zoubi et al[38] and Lendrum et al[39] recently documented successful conservative treatments in children younger than 12 years. One was staged as B1: lunate intact based on the amalgamation classification of Lichtman et al,[42] published in 2017.

Own Clinical Case

At the end of 2021, a 12-year-old right-handed boy was referred to our unit with a diagnosis of the Madelung deformity in the left wrist. The patient, with no history of trauma or other medical conditions, presented with chronic left wrist pain lasting for 3 months during wrist loading, though range of motion was normal and swelling was absent. Initial x-rays revealed a negative ulnar variance of 4 mm on the left and 3 mm on the right, without additional signs of the Madelung deformity. A removable wrist cast was prescribed pending further imaging. Two weeks later, a computed tomography (CT) scan confirmed lunatomalacia grade II according to the Lichtman classification in the left lunate (Fig. 9). Treatment involved immobilization in a thumb spica cast for 3 months. Follow-up CT scans after 4 months revealed progression to lunatomalacia grade IIIA, yet the patient's pain had significantly decreased. Based on the improved clinical symptoms, the spica cast was continued for 6 additional weeks, followed by 3 months of removable casting. Wrist exercises were prescribed 4 times daily, avoiding weight-bearing activities. A subsequent CT scan 4 months later showed complete restitution of the lunate. The cast was removed with recommendations to avoid heavy wrist loading for several weeks. One year after initiating conservative treatment, the patient was pain-free, with normal wrist range of motion and grip strength in both hands.

DISCUSSION

This systematic review aimed to identify the best treatment—surgical or conservative—for pediatric or juvenile lunatomalacia based on age, but no conclusive answer was possible. The main study limitations include small sample sizes, inconsistent data, varying patient ages, and publication bias favoring case reports. Despite these issues, this article is likely the most comprehensive collection of reported cases to date, identifying 86 cases worldwide, including our own. The study of Iwasaki et al[43] was excluded from our survey because of insufficient detailed information about the patients treated with osteotomies. The Spanish case report was also excluded due to the unavailability of the full text.[44] Most studies lack clarity on the parameters guiding treatment decisions. Authors often relied on comparable adult cases or personal experiences. The primary treatments include conservative plaster splinting or surgical interventions (eg, radial shortening or temporary joint fusion) aimed at unloading the lunate. However, it remains unclear when surgical therapy should replace conservative treatment. Of 10 patients younger than 15 years (Table 2), 5 treated conservatively had favorable outcomes. Among 4 surgically treated patients, 1 experienced complications (K-wire dislocation). This underscores the value of conservative splinting in patients younger than 15 years of age. Conservative cases involved casting for 9–30 weeks, whereas surgical cases included brief or no conservative trials. Publications with a conservative approach reported promising results, whereas surgical treatment often involved complications. Pain relief during splinting should guide clinical decisions. Our case illustrates that complete pain relief after 12 weeks of splinting can justify conservative management, even if radiological findings worsen initially. Although statistically proven recommendations were not possible due to limited and incomplete data, we advocate for nonsurgical treatment with regular follow-ups for patients younger than 15 years. Similarly, Lichtman et al[37] recommend avoiding surgery in this age group, emphasizing that evidence was limited.

Non-English publications[45,47–50] provided additional insights, but these were excluded from statistical analysis.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

PATIENT CONSENT

The patient provided informed consent.

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