Muscle and joint sequelae in brachial plexus injury

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Chapter 4

Serial casting for elbow flexion contractures in obstetric brachial plexus injury

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ABSTRACT

Background: The objective of this study was to evaluate the effectiveness of serial casting of elbow flexion contractures in obstetric brachial plexus injury.

Methods: A prospective consecutive cohort study was performed with a median follow-up of 5 years. Forty-one patients with elbow flexion contractures ≥30° were treated with serial casting until the contracture was ≤10°, for a maximum of 8 weeks. Range of motion, number of recurrences and patient satisfaction were recorded and analyzed using Wilcoxon signed-rank and Cox regression tests.

Results: Passive extension increased from a median of -40° (IQR -50 to -30) to -15° (IQR -10 to -20, $p < 0.001$). Twenty patients showed 37 recurrences. The baseline severity of passive elbow extension had a hazard ratio of 0.93 (95% CI 0.89 to 0.96, $p < 0.001$) for first recurrence. Median patient satisfaction was moderate. Four patients showed loss of flexion mobility and in two patients serial casting had to be prematurely replaced by night splinting due to complaints.

Conclusion: Serial casting improved elbow flexion contractures, although recurrences were frequent. The severity of elbow flexion contracture is a predictor of recurrence. We recommend more research on muscle degeneration and determinants involved in elbow flexion contractures to improve treatment strategies and prevent side-effects.

INTRODUCTION

Obstetric brachial plexus injury (OBPI) is caused by trauma to the brachial plexus during delivery, resulting in axonotmesis, neurotmesis and/or avulsion of some or all of the C5 through T1 nerve roots and/or trunks. The incidence varies between 0.4 and 4.6 per 1000 live births [1-3]. The natural course is diverse and depends on the extent and severity of the nerve lesions. Most children show full neurological recovery, but 20 to 35% have residual deficits for which reconstructive nerve surgery is indicated [4-8]. Despite improved function after natural recovery and nerve surgery, up to 35% of OBPI patients have residual muscle weakness, contractures and/or joint deformities and require additional treatments to further improve the function of the upper extremity, including soft-tissue releases, muscle tendon transfers and osteotomies [7, 9-11].

The main sequela of upper OBPI (C5-C6) at the elbow joint is flexion contracture, with a prevalence of 50 to 90%. In most cases, flexion contractures are limited to 10° to 30° and can be treated by range-of-motion exercising and nighttime splinting. In a minority of cases, however, the contracture exceeds 30°, and additional treatment is needed. An elbow flexion contracture greater than 30° has a severe impact on function in terms of the international classification of functioning, disability and health (ICF) activity and participation categories, since most daily living activities are executed in 30° to 130° of elbow flexion [12-14]. Literature on the management of elbow flexion contractures in patients with OBPI is scarce, and authors have proposed both non-surgical and surgical treatments, including splinting, serial casting, anterior elbow release and arthrodiatasis [14-19]. However, the studies all had limited patient numbers and lacked long-term follow up. Although serial casting is frequently applied and globally considered to be the preferred therapy, literature on the effect of stretching by serial casting for contractures is limited.

The primary objective of the current study was to evaluate whether serial casting of elbow flexion contractures would improve elbow extension in patients with OBPI. Secondary objectives were to assess the number of recurrences of elbow flexion contractures and to examine side-effects, including the effect of serial casting on biceps and triceps muscle strength and restrictions of flexion, supination and pronation. Patient satisfaction and predisposing patient characteristics influencing treatment outcome were also examined.

METHODS

A prospective consecutive cohort study was performed among children with OBPI treated with serial casting because of elbow flexion contracture.

Patients

Forty-one consecutive patients with OBPI and elbow flexion contracture of ≥30° were recruited at the outpatient clinic of the Leiden University Medical Center (LUMC). All recruited patients were enrolled in this prospective cohort study, as serial casting treatment was the standard treatment protocol for patients with ≥30° of flexion contracture (i.e. an extension deficit of 30°) despite regular physical therapy and nighttime splinting. Serial casting started with two weeks of casting in submaximal stretch to enable the child to get used to the cast and to allow for muscle accommodation. From the third week on, the serial casting was performed in maximal extension to maximize the effect of the casting period. Serial casting was only continued if the treatment was tolerated by the patients. This protocol remained unchanged throughout the inclusion period. Patients were excluded from this study if they had undergone prior surgery of the affected elbow, or if they had radial head dislocation. Patients were also excluded if the elbow flexion contracture benefitted the functional ability of the arm. For example, an elbow contracture can be functional in the case of poor hand function (Raimondi hand function scale scores below 3 [20]). All children and their parents were informed about common discomforts and functional burden during the casting period, like the inability to swim. They were also informed about side-effects like temporary restriction of active elbow flexion after cast removal.

All patients were treated by one staff member (R.G.H.H.N.), and serial casting was performed with weekly casting in maximal elbow extension, until the flexion contracture was 10° or less. Serial casting lasted for a maximum of 8 weeks. If the maximal extension was reached after serial casting, patients received a nighttime splint, using the final cast as a splint. Physical therapy was continued after serial casting, using range of motion exercises. The study protocol to prospectively evaluate children with OBPI was approved by the LUMC ethics committee.

Clinical parameters

The age, gender, affected side, severity of the lesion according to Narakas, type of primary treatment and age at nerve surgery were recorded [21]. All patients were evaluated at baseline, after serial casting (a median of 4 weeks, interquartile range (IQR) 4 to 6) and at annual regular follow-up, including passive elbow extension and flexion as well as passive forearm supination and pronation in 90° of elbow flexion. Elbow flexion and extension strengths were measured on a Medical Research Council (MRC) 0 - 5 scale, as muscle imbalance was considered one of the factors that can influence recurrence after treatment. Active elbow extension, flexion and forearm supination and pronation were assessed at follow-up to assess side-effects. We also recorded the number of weeks of serial casting, the number of recurrences and whether or not patients received additional treatment. Patient satisfaction regarding goal attainment was retrospectively assessed by an independent pediatric physiatrist, and critically checked by the first author, who subjectively examined the neurosurgeon's, orthopedic surgeon's and rehabilitation records, and rated patient satisfaction using a 5-point Likert scale: 1. very satisfied, 2. moderately satisfied, 3. neither satisfied nor dissatisfied, 4. moderately dissatisfied and 5. very dissatisfied. No patients were lost to follow-up, and baseline and follow-up range of motion data of all patients were available for analysis.

Statistical analysis

The Wilcoxon signed-rank test was used to test the statistical significance of clinical outcome parameters, as range of motion data was not normally distributed. Cox regression analysis was used to determine whether any of the clinical parameters were associated with recurrence of elbow flexion contractures, including age, gender, Narakas type, primary treatment, biceps and triceps muscle strength and number of weeks of serial casting. IBM SPSS Statistics version 20 was used for all statistical testing.

RESULTS

Elbow flexion contracture

Patient characteristics are shown in table I, showing separate data for each Narakas patient category. Clinical results are shown in table II, again showing separate data for each Narakas patient category, including passive extension, flexion, supination and pronation, number of patients with one or more recurrences, mean number of recurrences per patient, time until first recurrence, biceps and triceps muscle strength and patient satisfaction. The elbow range of motion for the total patient group is shown in figure 1. This total patient group had a median passive elbow extension of -40° (IQR -50 to -30) at the start of serial casting. After serial casting for a median of 4 weeks (IQR 4 to 6), the median passive extension had increased to -15° (IQR -10 to -20, $p < 0.001$). At follow-up, after an average of 5 years (IQR 3 to 6), the median passive extension was -25° (IQR -40 to -20, $p < 0.001$). During this period, 20 (49%) patients had a total of 37 recurrences of elbow flexion contracture ≥30° and were treated a second time with serial casting, following the same procedure. The median time to the first recurrence was 2 years (IQR 1 to 4). Excluding patients with recurrent elbow flexion contracture, the median passive extension was increased from -30° (IQR -40 to -30) at baseline to -20° (IQR

-25 to -20, $p < 0.001$) at follow-up. One patient had 7 recurrences of elbow flexion contractures and was treated with multiple serial castings. At an age of 5 years, this patient underwent anterior capsule release because of an elbow flexion contracture of 70°. After 12 years of follow-up, the patient's elbow flexion contracture was 50°. Patient satisfaction was scored at a median value of 2, indicating 'moderately satisfied' (IQR 1 `very satisfied´ to 2 ´moderately satisfied´).

Side-effects / complications: elbow flexion, supination and pronation

Two participants scored 5 on the satisfaction scale, indicating 'very dissatisfied' as regards goal attainment, in both cases due to loss of elbow flexion. Two participants reported greater than expected discomfort in the first week of serial casting, including pain and hand edema. In both of these children, the serial casting had to be prematurely replaced by splinting as a result of complaints. The median baseline passive elbow flexion was 150° (IQR 145 to 150). After serial casting for

Table I: Patient characteristics

Narakas type	C5 – C6 N = 11	C5 – C7 N = 21	C5 – T1 N = 9
Sex (male)	5	8	6
Age at baseline measurement # * (years)	6.8 (4.71)	7.1 (4.36)	10.9 (4.68)
Side (right / left)	7 / 4	10 / 11	4 / 5
Age at primary treatment § * (years)	0.5 (0.26)	0.5 (0.17)	0.4 (0.24)
Conservative / neurolysis / nerve transplantation	8 / 1 / 2	6 / 1 / 14	1 / 0 / 8
Anterior division of superior trunk	2	10	8
Posterior division of superior trunk	2	9	7
Medial trunk	1	1	5
Suprascapular nerve	2	9	6
Secondary procedures			
Shoulder external rotation surgery	3	9	3
Pronation osteotomy of the ulna and/or radius	0	4	2
Wrist extension surgery	0	4	3
Botulinum toxin injection subscapularis	1	0	0
Botulinum toxin injection triceps	0	0	1
Botulinum toxin injection biceps and brachialis †	0	1	0
Biceps rerouting surgery †	0	2	1
Elbow manipulation in narcosis †	1	0	0
Anterior capsule release elbow †	1	0	0
Casting ‡ (weeks)	6 (3 to 7)	4 (3 to 5)	6 (4 to 6)
Follow-up * (years)	5.4 (4.15)	5.6 (3.03)	3.0 (2.97)

#This is the age at the start of serial casting treatment. *The values are given as mean with standard deviation. §This is the age at neurosurgical intervention. †Secondary procedure performed after inclusion. ‡The values are given as median and interquartile range.

a median of 4 weeks (IQR 4 to 6), the median passive flexion had decreased to 140° (IQR 120 to 150, $p = 0.001$) and at a mean follow-up of 5.0 years (SD 3.29) it had increased again to 145° (IQR 140 to 150, $p = 0.006$) as shown in figure 1. Four patients showed a loss of elbow flexion of 50° or more. One child with a severe decrease in elbow flexion, to 85°, had to be treated two times with manipulation under general anesthesia, after which the elbow flexion stabilized at 130°. During follow-up, six patients were treated with pronation osteotomy of the ulna and/or radius, including three patients who were additionally treated with biceps tendon transfer at a mean of 5.3 years (SD 1.05) after the start of serial casting. However, these supination contractures had no relation with the serial casting. When these six patients are excluded, there was no change in the median passive supination at follow-up ($p = 0.13$). The median baseline passive pronation was 90° (IQR 30 to 90) and had not changed either after serial casting or at the final follow-up. The median

Table II: Results of serial casting treatment

Narakas type	C5 – C6 N = 11	C5 – C7 N = 21	C5 – T1 N = 9
Passive extension ‡ (°)			
At baseline	-30 (-40 to -30)	-40 (-48 to -30)	-45 (-50 to -38)
After 4 weeks	-20 (-20 to -14)	-15 (-20 to -10)	-13 (-20 to -6)
At follow-up	-25 (-30 to -20)	-25 (-40 to -20)	-30 (-40 to -18)
Passive flexion ‡ (°)			
At baseline	150 (145 to 150)	150 (145 to 150)	150 (143 to 150)
After 4 weeks	143 (101 to 150)	140 (120 to 150)	140 (100 to 150)
At follow-up	143 (130 to 150)	145 (140 to 150)	145 (140 to 150)
Passive supination ‡ (°)			
At baseline	90 (88 to 90)	90 (74 to 90)	90 (85 to 100)
After 4 weeks	90 (90 to 90)	90 (90 to 90)	90 (90 to 96)
At follow-up	90 (78 to 90)	70 (30 to 90)	90 (10 to 90)
Passive pronation ‡ (°)			
At baseline	90 (80 to 90)	90 (38 to 90)	30 (5 to 90)
After 4 weeks	90 (90 to 90)	65 (39 to 83)	10 (-11 to 90)
At follow-up	90 (80 to 90)	80 (60 to 90)	70 (0 to 90)
Patients with recurrence	4 (36%)	10 (48%)	6 (67%)
Recurrences per patient #*	3 (3.0)	2 (1.0)	2 (0.8)
Time of first recurrence * (years)	2.9 (2.02)	2.2 (1.85)	2.3 (1.08)
Biceps muscle strength ‡ (MRC)	4 (4 to 4)	4 (4 to 5)	4 (3 to 4)
Triceps muscle strength ‡ (MRC)	5 (5 to 5)	4 (3 to 5)	4 (3 to 4)
Patient satisfaction ‡	2 (1 to 3)	2 (1 to 2)	2 (2 to 3)

‡The values are given as median and interquartile range. # The mean number of recurrences of elbow flexion contracture per patient after the initial serial casting treatment. *The values are given as mean with standard deviation.

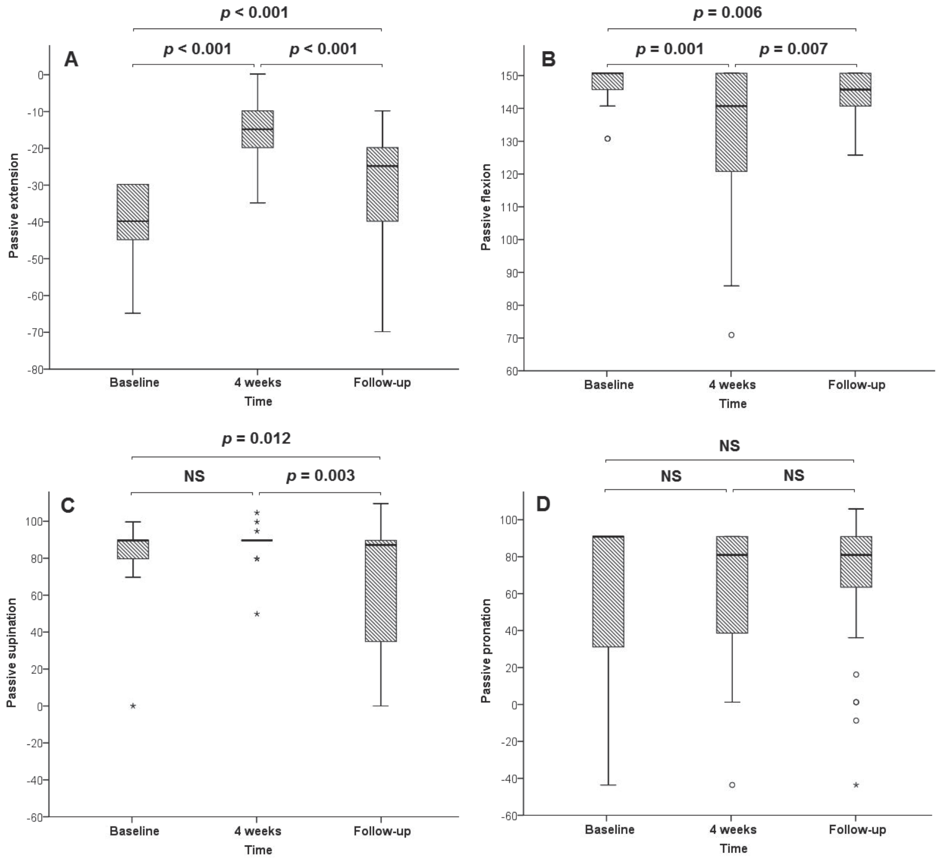

Figure 1: Clinical results
Clinical results presented as box plots of passive extension (A), flexion (B), supination (C) and pronation (D) at baseline, after a median of 4 weeks of serial casting and at final follow-up after 4.6 years. Wilcoxon signed-rank *p*-values are shown for changes between baseline measurement, measurement after a median of 4 weeks of serial casting, and measurements at follow-up. NS = not significant.

biceps muscle strength in the total patient group was MRC 4 (IQR 4 to 4) and the median triceps muscle strength was MRC 4 (IQR 3 to 5), both of which did not change during follow-up. As expected, the median triceps muscle strength was lower in patients with C7 involvement (MRC 4, IQR 3 to 5) than in patients with a C5-C6 lesion (MRC 5, IQR 5 to 5, $p = 0.02$). The median biceps muscle strength (MRC 4, IQR 4 to 4) was lower than the median triceps muscle strength (5, IQR 5 to 5) in patients with a C5-C6 lesion ($p = 0.025$). Biceps and triceps muscle strengths were comparable in patients with a C7 involvement ($p = 0.34$). The median passive elbow extension at baseline was lower in patients with C7 involvement (-40°, IQR

-50 to -30) than in those with a C5-C6 lesion (-30°, IQR -40 to -30). This difference approached significance ($p = 0.07$). At follow-up, the median active extension was -30° (IQR -40 to -20), median active flexion 140° (IQR 135 to 145), median active supination 70° (IQR 10 to 90) and median active pronation 80° (IQR 40 to 85).

Predictors of recurrent elbow flexion contractures

Baseline median passive extension was -45° (IQR -60 to -30) in patients with recurrence of elbow flexion contracture, compared to -30° (IQR -40 to -30) in patients without recurrence, but this difference did not reach statistical significance ($p = 0.16$). Patients whose flexion contracture recurred showed no significant difference in passive flexion, supination or pronation at baseline or after serial casting, compared to patients without recurrences. However, their median extension decreased further during follow-up, despite further serial casting treatments, from -20° (IQR -20 to -10) to -40° (IQR -50 to -30, $p = 0.001$). At follow-up, patients with and without recurrence of flexion contracture did not differ in terms of passive or active flexion, supination or pronation, or biceps or triceps muscle strength.

Multivariate cox regression was used to model the time to first recurrence of elbow flexion contracture. Severity of passive elbow extension at baseline had a hazard ratio of 0.93 (95% CI 0.89 to 0.96, $p < 0.001$) for first recurrence of elbow flexion contracture. The following factors were not associated with recurrence of elbow flexion contracture: age, gender, Narakas type, primary treatment, biceps or triceps muscle strength and number of weeks of serial casting.

DISCUSSION

In the present study, serial casting of elbow flexion contractures in children with OBPI improved passive elbow extension, although recurrence of elbow flexion contractures was frequently observed (49%). Passive elbow flexion was decreased after serial casting. Four cases of a severe decrease in elbow flexion (i.e. more than 50° loss of elbow flexion) were encountered after casting, for which one patient required manipulation under general anesthesia. Although all patients in this population recovered during follow-up, the severe decrease in elbow flexion should be interpreted as a serious side effect. Passive supination and pronation were not affected by serial casting, and no changes were observed in biceps or triceps muscle strength. The more severe the contracture was at the start of the serial casting, the higher the likelihood of recurrence.

In terms of the ICF activity and participation levels, elbow flexion contractures are generally associated with functional hand positioning limitations, for example during balancing, cycling, or activities like carrying a bag or leaning on a bar. Literature on functional issues is scarce, and many questions remain about the clinical relevance of treating elbow flexion contractures. A study by Morrey demonstrated that most daily living activities are executed in the 30° to 130° range of motion of elbow flexion [13], and skills such as carrying a bag require even more elbow extension. These arguments prompted us to treat patients with persistent elbow flexion contractures ≥30° with serial casting. Treatment goals in our study also related to cosmetic issues. Patients with elbow flexion contractures below 30° are generally treated with physical therapy and night splinting only.

Literature on the treatment of elbow flexion contractures ≥30° is limited. In a recent study by Sheffler et al., 9 patients with elbow flexion contractures ≥30° were treated with serial casting [14]. They showed an improvement in passive elbow extension from -49° to -30°, which is less favorable than the results in the present study, which found an improvement in passive elbow extension from -40° to -15°. The more favorable outcome in our population (passive elbow extension -15° versus -30°) could be the result of an earlier start of the serial casting and the duration of serial casting, which was not reported by Sheffler et al. [14]. These authors found a further deterioration of the elbow flexion contracture of 4.4% a year during 27 months of follow-up, which is consistent with our finding of 10° of deterioration over 5 years of follow-up. Ho et al. treated 19 patients non-surgically for elbow flexion contractures, and found an improvement in passive elbow extension from -48° to -17°. However, their results included patients treated with serial casting as well as those treated with splinting [17].

The etiology of flexion contracture of the elbow in children with OBPI is unknown and needs further elucidation [10]. Current hypotheses in the literature include reinnervation of the elbow flexors prior to that of the elbow extensors, leading to muscle imbalance, and/or co-contraction as a cause of elbow flexion contractures [12]. Our study found no differences in biceps and triceps muscle strength of patients with lesions of both C5-C6 and C7. Patients with lesions only of C5-C6 developed an elbow flexion contracture despite stronger triceps than biceps muscles. Thus, the results of our study do not support muscle strength imbalance as the cause of elbow flexion contractures. Other hypotheses include changes in the partially denervated muscle itself as a cause of elbow flexion contracture, including muscle atrophy, fattening and fibrosis [22]. It is also possible that elbow flexion contractures could be caused by positional preferences of the arm in brachial plexus injury. A mild elbow flexion contracture could facilitate the weak elbow flexors, as limitation of elbow flexion leads to greater impairment of daily activities [13]. This, in conjunction with the internal rotation contracture present in the shoulder, attenuates the flexion position of the elbow even further.

Strengths of this study include that it is the largest case series so far on the treatment outcome of OBPI patients with severe elbow flexion contractures. We present clinical outcomes and complication rates, giving further clues about the etiology of elbow flexion contractures. To our knowledge there have not been any reports of serial casting studies using a 5-year follow-up. A limitation of this study is the lack of control groups to compare different treatment modalities, making it a prospective consecutive cohort study. The global use of the therapy and its presumed effect make it difficult to obtain ethical approval for including control groups and to motivate parents to participate in a randomized controlled trial. One of our messages is that the effect should not be overestimated, and we strongly recommend that randomized controlled trials be organized. Another limitation is that therapy compliance regarding night splinting and co-interventions like physical therapy were not recorded, which might have influenced recurrences of elbow flexion contracture. A third limitation concerns a lack of reproducibility as a result of individual variation in therapy. The maximum stretch force applied during serial casting depends on the interaction with and tolerance of the child and/or parent and therefore might have influenced the outcome of the serial casting treatment. On the other hand, a strength of this study is that the intervention performed was usual care, and therapy did not change because of participation in the study. This was also the reason why we had not expected the dissatisfaction that we found, and we therefore had to assess patient satisfaction retrospectively. Future studies should assess the patient satisfaction and goal attainment prospectively.

In conclusion, this study demonstrates that serial casting can improve elbow flexion contractures in children with OBPI. Complications include temporary decrease of elbow flexion and a high rate of recurrence of elbow flexion contractures, stressing the importance of close follow-up. The severity of the elbow flexion contracture at the start of serial casting is a predictor of recurrence of the elbow flexion contracture. Further research into the etiology and treatment strategies could optimize the management of elbow flexion contractures in OBPI. This could include studies on the many recent developments in dynamic splinting techniques. As including an untreated control group is hardly feasible, we recommend randomized controlled trials comparing generally accepted therapy strategies such as serial casting and dynamic splinting. More research is needed to evaluate serial casting of elbow flexion contractures at the ICF level of activity and participation. In order to prevent side-effects, we recommend a close follow-up, careful physical examination during the cast changes and critical estimation of the casting period, which should be as short as possible.

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