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Plastic and Reconstructive Surgery Advance Online Article

DOI: 10.1097/PRS.00000000000011877

Spontaneous recovery of active shoulder external rotation in patients with brachial plexus birth injury

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Conflicts of interest: The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Financial disclosure statement: The authors received no financial support for the research, authorship, and/or publication of this article.

Short running head: Brachial plexus birth injury

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Abstract

Background: Brachial plexus birth injuries (BPBI) occur as a result of a difficult delivery. External rotation of shoulder (ER) is usually one of the last movements which may recover. There is no consensus about the predicting factors for spontaneous recovery or the optimal timing for surgical treatment of ER in BPBI patients. The aim of our retrospective study was to describe spontaneous recovery of active ER and evaluate predicting factors for the recovery.

Methods: We screened 562 patients and identified a consecutive cohort of 103 BPBI patients, who had no active ER at the age of 3 months. We systematically collected clinical data on recovery. In addition, we assessed whether early recovery of elbow flexion, shoulder abduction or Narakas grade at 1 month predicts ER recovery.

Results: Fifty-two (51%) patients spontaneously recovered ER, 44% of whom were recovered by the age of 1 year, 83% by 1.5 years, 92% by 2 years and 98% by 3 years. A breakpoint in the slope of the curve showing proportion of recovered patients occurred at 2 years of age. Recovery of active ER was significantly associated with early elbow flexion and Narakas grade at 1 month, but not with early active shoulder abduction.

Conclusions: Most spontaneous recovery of ER in patients with BPBI occurs until 2 years of age, which thus can be considered a meaningful follow-up period for spontaneous recovery of ER. This information should be considered when making decision about peripheral nerve transfer surgery to improve ER in BPBI.

Introduction

Brachial plexus birth injuries (BPBI) are caused by traction during a difficult delivery. Incidence of BPBI varies from 0.9 to 5.1 per 1000 births¹⁻³ and BPBI usually affects the C5 and C6 roots.

⁴ Of all the BPBI patients, 66–92 % recover completely^{1,5,6}. Based on the regeneration speed of axons, the time for complete neurological recovery can be up to 3 years, while ongoing functional recovery is rarely reported after 3 years of age⁵.

It's commonly suggested that children who recover anti-gravity elbow flexion function later than 3 months of age, are more likely to have worse shoulder function than those who recover elbow flexion earlier⁶⁻⁸. Accordingly, absent elbow flexion function at 3 months of age is generally regarded to reflect insufficient spontaneous nerve recovery and to be an indication for early surgical brachial plexus reconstruction^{5,9,10}. However, elbow flexion often recovers early, while active external rotation of shoulder (ER) is among the last movements to recover in BPBI¹. The probability of spontaneous recovery of ER over time is not known, and thus, there is no consensus on the optimal timing for making treatment decision about a possible distal nerve transfer surgery (spinal accessory nerve to suprascapular nerve, SAN-SSN) to activate ER.¹⁹⁻
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The rationale for the study was to evaluate spontaneous recovery of active ER after BPBI. In addition, we assessed whether early elbow flexion, Narakas grade at 1 month and shoulder abduction are predictors for spontaneous recovery of active ER. These results can be used when making treatment decision about surgical treatment for absent active ER.

Methods

In this retrospective study, we evaluated spontaneous recovery of active shoulder ER in BPBI patients who had no ER at the age of 3 months. The study conforms to the research guidelines of [study hospital] Ethical Review Board. The study results are reported according to STROBE statement¹¹. To identify all potentially eligible patients, we screened children born between

years 2000 and 2021 from the [study hospital] electronic medical record with the diagnosis codes for birth injuries to peripheral nervous system (ICD-10: P14.0, P14.1, P14.2, P14.3, P14.8, P14.9). From those ($n=562$) we identified the patients who met the inclusion criteria of the study: 1) patient had confirmed BPBI diagnosis, 2) had not fully recovered (full recovery was defined as recovery of all upper extremity functions to grade 7 on the active movement scale (AMS))¹² within the first 3 months of life, 3) ER had been absent (AMS 0–5) at the clinical evaluation after 3 months of age, 4) patient's first clinical evaluation at the study hospital had been before 3 years of age and 5) duration of follow-up for possible recovery was minimum 3 years. Patients who had underwent brachial plexus reconstruction or SAN-SSN before 9 months of age were excluded, because these operations prevent spontaneous recovery of ER. (Figure 1) The data on patient and injury details and clinical findings during follow-up were collected from patient record using Last Observation Carried Forward (LOCF) because of some missing follow-up observations¹³.

The AMS is an eight-grade scale for evaluating motor function and it can be used with children of any age. The scale range is from 0 to 7, where AMS 0 means no muscle contraction, AMS 5 indicates active movement less than half range of movement against gravity, AMS 6 active movement at least half range of movement against gravity and AMS 7 normal active movement.

¹² In our study, active shoulder ER, elbow flexion and shoulder abduction at different time points were translated into AMS grade during the data collection. When the patient achieved ER AMS 6, in which the forearm reaches sagittal plane (neutral position) when arm in full adduction and elbow in 90 degrees flexion, the ER function was considered as recovered.

Extent of BPBI was described with Narakas classification at the age of 1 month¹⁴. Evaluation of ER at follow-up assessments was performed and recorded independently by a hand surgeon specialized in BPBI and a pediatric physiotherapist. Passive and active ER were evaluated in full shoulder adduction and elbow in 90 degrees of flexion. The presence of active ER was evaluated

at the ages of 3–4 months, 5–6 months, 8–10 months, 1 year, 1.5 years, 1.75 years, 2 years, 2.25 years, 3 years, 5 years and 7 years. Last evaluated follow-up visit took place at a mean age of 12.6 years (SD 2.4). The age when active ER improvement to AMS 6 was first observed was determined for every patient if such recovery occurred.

Physiotherapy protocol used in our hospital consists of passive exercises started in few days after birth, sensory re-education and progressive strengthening of active movements. It consists of weekly appointment with a physiotherapist and free passive range of motion exercises, particularly into shoulder abduction and external rotation, at home according to the instructions given by the physiotherapist.

In our treatment protocol, patients with absent antigravity elbow flexion at 3 months are referred to surgical exploration of brachial plexus. If there are viable nerve roots, reconstruction of brachial plexus with interposition nerve grafts is indicated. In case of upper root avulsions, nerve transfers are used, such as SAN–SSN for ER. If early surgical treatment is not indicated and there is persistent deficiency in ER, we consider 1.5 years as a threshold for making decision about SAN–SSN surgery. Tendon transfers, e.g. latissimus dorsi and teres major tendon transfer to infraspinatus tendon, have been used as later stage interventions in patients with absence of shoulder function, but nowadays they are seldom used due to SAN–SSN technique which is the current standard practice.

All the patients included in the follow-up of spontaneous recovery had at least 70 degrees of passive ER. Botulinum toxin-A (BTX) injections to shoulder internal rotator muscles have been used at the study hospital since year 2006. BTX injections are effective in maintaining the sufficient passive movement and shoulder congruency¹⁵, and were administered to study patients if restriction of passive ER was observed before 1.5 years of age while waiting for the spontaneous recovery of active ER. Thus, patients treated in the beginning of the study period before the use of BTX injections were more likely to develop internal rotation contractures.

Patients with passive restriction of ER or subluxation of humeral head observed after 1.5 years of age are treated with anterior release surgery and subscapular tendon lengthening, if needed.

Patients who underwent an anterior release surgery during the study period, were separated from the follow-up of spontaneous recovery.

Operative treatment for active ER (SAN–SSN or a tendon transfer surgery) prevented further spontaneous recovery of the nerve and/or made evaluation of spontaneous recovery of ER impossible. Patients who underwent one of these procedures were identified and separated from the follow-up group at the time of the operation. First SAN–SSN surgery in our patient cohort took place in year 2011, and before that, if surgical treatment for active ER was indicated, patients were treated with tendon transfers.

A segmented linear regression analysis was conducted to find a transition point where a remarkable change happens in the slope of the curve representing cumulative percentage of recovered patients. As a planned sensitivity analysis, we evaluated the impact of estimation method for time point for recovery of ER to AMS 6. For that analysis, we identified and used the last clinical evaluation when ER AMS 6 was not yet observed in every recovered patient, because the true time point for recovery of ER to AMS 6 was between the last observation when AMS was still < 6 and the next when AMS 6 was observed.

To evaluate the prognostic value of early elbow flexion and shoulder abduction when estimating recovery of ER, we divided the patients into groups based to their early elbow flexion (Group EF 1: AMS 6–7 before 4 months of age and Group EF 2: AMS 0–5 at 4 months of age) and shoulder abduction (Groups ABD 1 and ABD 2, respectively). Four months was used as the age limit, because we wanted to include the finding from clinical appointment at the age of 3 months, which in practice may take place 1–2 weeks after the precise age of 3 months. For the analysis regarding association between Narakas classification at 1 month and recovery of ER, we pooled Narakas 3 and 4 groups as there was only one patient in group 4. To assess the impact

of elbow flexion, shoulder abduction and Narakas classification on recovery of ER, we used Cox proportional hazards model. The patients who had undergone any surgery which may affect ER during the follow-up period, or did not recover active ER, were marked as censored observations.

For statistical analyses we used R, version 4.3.2 (R Foundation for Statistical Computing, Vienna, Austria) (package ‘segmented’¹⁶). Ages and recovery times are presented as mean with standard deviation (SD). The estimated breakpoint in the curve of recovered patients is reported with 95% confidence interval (CI) and coefficient before and after the breakpoint with standard error (SE), 95% CI for the coefficient and associated *p*-value. The proportional hazards assumption was tested with Schoenfeld residuals test, and crude and adjusted hazard ratios (CHRs and AHRs) for the Cox proportional hazards regression are presented with 95% CIs and *p*-values in Supplemental Digital Content of the article (Table, Supplemental Digital Content 1). ANOVA *p*-values are presented for each variable in the model. Kaplan-Meier hazard curves are shown with *p*-values from log-rank tests. *P*-values < 0.050 were considered statistically significant.

Results

We identified 103 patients born between years 2000 and 2021 with BPBI and missing active ER at 3 months of age. Patient characteristics are shown in Table 1.

Recovery of ER

Fifty-two (51%) out of 103 patients spontaneously recovered active ER (Figure 2). Of the 52 that recovered ER, 23 (44%) recovered by 1 year of age, 43 (83%) by 1.5 years, 48 (92%) by 2 years, and 51 (98%) by 3 years. The most significant breakpoint in the curve depicting the cumulative proportion of patients with spontaneous ER recovery was at the age of 22 months. Segmented regression analysis is presented in Table 2. In the confirmatory sensitivity analysis

with differently defined time points for recovery, 45 (87%) of 52 patients would have been recovered by 1 year of age, 48 (92%) by 1.5 years and 52 (100%) by 2 years.

Twenty-one patients underwent SAN-SSN (at a mean age of 22 months, SD 12) and two underwent SAN-SSN and anterior release with subscapular tendon lengthening at the same procedure at 14 and 18 months. Two patients underwent a tendon transfer at 42 and 92 months, and ten patients a tendon transfer with anterior release and possible subscapular tendon lengthening (mean 69 months, SD 19). Fourteen patients, of whom 12 developed dorsal subluxation of humeral head and glenohumeral dysplasia, ended up in an anterior release surgery after persistent restriction of passive ER (mean age 33 months, SD 21) without any simultaneous surgery for active ER. (Table 1) Eight of them recovered active ER after the release procedure suggesting that without passive restriction the total proportion of patients with spontaneous recovery of active ER would have been bigger.

Two patients did not recover active ER to AMS 6 and did not undergo any surgical treatment. Both of these patients had restriction of passive ER already in the first months of life. In six patients who spontaneously recovered ER to AMS 6 we observed deterioration of ER during the follow-up, at a mean age of 4.8 years (SD 3.8). Two of these patients developed dorsal subluxation of humeral head and glenohumeral dysplasia, with one experiencing permanent deterioration leading to AMS 0 ER. Five patients regained active ER AMS 6 after a period of weaker function.

Relationship between early elbow flexion and recovery of ER

Patients were divided into two groups based on their elbow flexion at 4 months: AMS 6–7 (group EF1) and AMS 0–5 (group EF2). Eight patients were excluded from this part of the analysis due to missing information regarding elbow flexion. (Table 1) Sixty-two percent (28/45) of patients in group EF1 and 46% (23/50) in group EF2 spontaneously recovered active

ER AMS 6. The groups EF1 and EF2 were significantly different in Kaplan-Meier analysis and univariate Cox regression (Figure 3, Table, Supplemental Digital Content 1).

Relationship between early shoulder abduction and recovery of ER

Patients were divided into two groups based on their shoulder abduction at 4 months: AMS 6–7 (group ABD1) and AMS 0–5 (group ABD2). Thirty-three patients had missing information regarding shoulder abduction before 4 months. (Table 1) Sixty-seven percent (10/15) of patients in group ABD1 and 53% (29/55) in group ABD2 spontaneously recovered ER AMS 6. There was no difference between the groups ABD1 and ABD2 in the survival analysis (Figure 4, Table, Supplemental Digital Content 1).

Relationship between Narakas classification at 1 month and recovery of ER

Seventy-one percent (20/28) of patients in group Narakas 1, 41% (21/51) in group Narakas 2 and 67% (6/9) in group Narakas 3–4 spontaneously recovered active ER. Information regarding Narakas grade at 1 month was missing for 15 patients (Table 1). The groups Narakas 1 and 2 were significantly different in Kaplan-Meier analysis and univariate and multivariate Cox regression models (Figure 5, Table, Supplemental Digital Content 1).

Discussion

In our consecutive cohort, 51% of the BPBI patients with absent ER still present at three months of age spontaneously recovered good active ER. There was a decline in the rate of spontaneous ER recovery at 2 years of age, after which spontaneous recovery of ER was not likely. Two years of age can thus be considered as meaningful upper limit for follow-up of spontaneous recovery of ER. Early active elbow flexion was associated with spontaneous recovery of active ER, but early active abduction of shoulder was not. Our findings suggest that in patients with no indication for early surgical treatment before age of 6 months, spontaneous recovery of ER can be expected up to 2 years, but when considering the optimal timing for decision about possible peripheral nerve transfer surgery, the relationship between muscle denervation time and surgical outcome must also be taken into account.

The strength of this study is a large unselected consecutive patient cohort with good adherence to long term follow-up. Some studies about conservative treatment outcomes exist^{1,4,5,7,8,17-21}, but to our knowledge, there are no studies about schedule of spontaneous recovery of ER in BPBI patients. However, a retrospective study design, heterogenic patient cohort, small number of certain injury types, and some missing information made it difficult to compare the spontaneous recovery outcomes based on the type of injury. Further, the type and grade of BPBI – description of the affected roots and classification of injury to axonotmesis, neurotmesis or root avulsion – was not included in the analysis, because determination of these can be very difficult and inaccurate, particularly without exploration in conservatively treated patients^{5,22}. Secondly, examination and grading of individual muscle activity in young children can be difficult. Evaluations were based on a visual estimation, which is the only feasible practice. To reduce inaccuracy in the measurements, we used both physician's and physiotherapist's independent assessments and records. Thirdly, as the true time point for recovery to AMS 6 usually took place between clinical evaluations, there was some inaccuracy in the time points for recovery to AMS 6 and this translates into later time point of recovery of active movement. However, based on our sensitivity analysis, which had the maximum opposite (early recovery) bias with time points, we confirmed that method for estimating the time point for ER recovery to AMS 6 does not substantially influence the results.

When our results are compared with previous reports of complete spontaneous recovery of ER after BPBI^{1,5,6}, the proportion of patients who recovered active ER was slightly worse in our cohort of 103 patients. This is probably because patients with a minor injury and full recovery of ER within the first three months of life were excluded from our study. If the patients with early spontaneous recovery are also considered, spontaneous recovery of ER to AMS 6 was observed in 73%, which is in line with the previous reports.

There is no consensus on the optimal timing for making the decision about – or performing – surgical procedures for those BPBI patients who do not spontaneously recover ER^{9,10,23}. Our results show that spontaneous recovery of ER can be expected until the age of 2 years. It must be noted that the patients in our study with recovery of ER after 2 years were evaluated during the beginning of our study period when SAN-SSN transfer surgery had not yet become an established practice and thus the systematic evaluation of shoulder muscle functions was performed less systematically during the early years. In addition, the use of BTX injections has enabled maintaining passive ER and shoulder congruency while waiting for the recovery of active movement. Early recognition and treatment of possible restriction of passive range of shoulder movement seems important to provide for unobstructed recovery of active ER¹⁵. Morphological defects progress over time if the muscle forces are absent^{22,24,25} and glenohumeral joint incongruence can be detected as early as at 2 months of age^{22,25}. In turn, deterioration of ER function in some patients could have been due to insufficient recovery of the nerve, which may lead to inadequate development of affected muscles and joint structures when the upper extremity grows in size. The majority of these patients had initially recovered AMS 6 ER after 1 year of age.

In our study, early elbow flexion function significantly associated with recovery of active ER, which is in agreement with previous reports by Waters and Smith et al.^{8,18}. In contrast, Xu et al. reported that none of the patients with no elbow flexion at 3 months of age recovered functional active ER, but their retrospective study included only patients who had already been treated in other hospitals for 3 to 4 years with no recovery¹⁹. On the contrary, early active shoulder abduction did not correlate with recovery of active ER, which has also been suggested by Michelow et al.⁷. Patients with Narakas grade 1 at 1 month recovered ER more often when compared with patients with Narakas grade 2. Due to small number of patients with Narakas grade 3 or 4, no further conclusions can be drawn about their recovery compared with the other

groups. Nevertheless, functional recovery of BPBI patients may be incorrectly predicted if only one prognostic parameter is used ⁷.

Lack of standard outcome variables has led to a variety of different outcomes in studies on BPBI patients and that prevents pooling and comparing specific outcomes from different studies. Furthermore, pooled results can be misleading particularly because of differences in patient referral indications, age at the first clinical evaluation, and definitions or determination of reported variables ¹⁴. Further research is needed on recovery of shoulder function in BPBI patients to determine the optimal combination of prognostic factors for spontaneous recovery of ER and on the relationship between timing of possible peripheral nerve transfer (SAN-SSN) and its outcome. This would help to determine the optimal age for making decision about possible peripheral nerve transfer within the 2-year window of expected spontaneous recovery of

ER.References

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Figure legends

Figure 1. Flow diagram for inclusion and exclusion of patients.

Figure 2. Recovery of active shoulder external rotation (ER) in 103 patients. Area in chart is showing cumulative percentage of patients that recovered ER spontaneously to Active Movement Scale (AMS) 6. Patients were excluded from the follow-up group if they underwent any of the following procedures: an anterior release surgery with subscapular tendon lengthening if needed due to restricted passive ER, spinal accessory nerve to suprascapular nerve transfer (SAN-SSN) with or without simultaneous anterior release and subscapular tendon lengthening if needed, and a tendon transfer surgery with or without simultaneous anterior release and subscapular tendon lengthening if needed.

Figure 3. Kaplan-Meier hazard curves and log-rank test p -value for active elbow flexion as a predictor for active shoulder external rotation (ER). Blue indicates the accumulated probability of ER recovery in patients with Active Movement Scale (AMS) 6–7 elbow flexion before 4 months of age and red indicates the accumulated probability of ER recovery in patients with AMS 0–5 elbow flexion at 4 months of age. Patients that are censored either did not achieve recovery of ER to AMS 6 by the end of the follow-up or underwent any of the following procedures: an anterior release surgery with subscapular tendon lengthening if needed due to restricted passive ER, spinal accessory nerve to suprascapular nerve transfer (SAN-SSN) with or without simultaneous anterior release and subscapular tendon lengthening if needed, and a tendon transfer surgery with or without simultaneous anterior release and subscapular tendon lengthening if needed.

Figure 4. Kaplan-Meier hazard curves and log-rank test p -value for active shoulder abduction as a predictor for active shoulder external rotation (ER). Blue indicates the accumulated probability of ER recovery in patients with Active Movement Scale (AMS) 6–7 shoulder abduction before 4 months of age and red indicates the accumulated probability of ER recovery in patients with

AMS 0–5 shoulder abduction at 4 months of age. Patients that are censored either did not achieve recovery of ER to AMS 6 by the end of the follow-up or underwent any of the following procedures: an anterior release surgery with subscapular tendon lengthening if needed due to restricted passive ER, spinal accessory nerve to suprascapular nerve transfer (SAN-SSN) with or without simultaneous anterior release and subscapular tendon lengthening if needed, and a tendon transfer surgery with or without simultaneous anterior release and subscapular tendon lengthening if needed.

Figure 5. Kaplan-Meier hazard curves and log-rank test p -value for Narakas grade at 1 month¹⁴ as a predictor for active shoulder external rotation (ER). Patients that are censored either did not achieve recovery of ER to AMS 6 by the end of the follow-up or underwent any of the following procedures: an anterior release surgery with subscapular tendon lengthening if needed due to restricted passive ER, spinal accessory nerve to suprascapular nerve transfer (SAN-SSN) with or without simultaneous anterior release and subscapular tendon lengthening if needed, and a tendon transfer surgery with or without simultaneous anterior release and subscapular tendon lengthening if needed.

Table, Supplemental Digital Content 1. Cox proportional hazards regression for spontaneous recovery of shoulder external rotation (ER).

Table 1. Patient characteristics (number of patients).

Gender		
	Female	54
	Male	49
Affected side		
	Right	58
	Left	45
Narakas classification at 1 month		
	Narakas 1 (C5–C6)	28
	Narakas 2 (C5–C6–C7)	51
	Narakas 3 (C5–C6–C7–C8–TH1)	8
	Narakas 4 (C5–C6–C7–C8–TH1 + Horner)	1
	Not available	15
Elbow flexion at 4 months		
	AMS 0–5	50
	AMS 6–7	45
	Not available	8
Shoulder abduction at 4 months		
	AMS 0–5	55
	AMS 6–7	15
	Not available	33
Surgical treatment for ER		
	SAN–SSN	23
	Tendon transfer for ER	12
	Anterior release (all)	26
	Humeral rotational osteotomy	4

AMS = Active Movement Scale; SAN–SSN = spinal accessory nerve to suprascapular nerve transfer; ER = active shoulder external rotation

Table 2. Segmented linear regression for spontaneous recovery of active shoulder external rotation (ER).

Segment	Breakpoint (months)	95% CI (breakpoint)	Coefficient	SE	95% CI (coefficient)	<i>p</i>-value
Before 22.36	–	–	4.81	0.13	4.54–5.09	<0.001*
After 22.36	22.36	21.61–23.12	0.31	0.04	0.23–0.38	<0.001*

CI = confidence interval; SE = standard error.

Figure 1

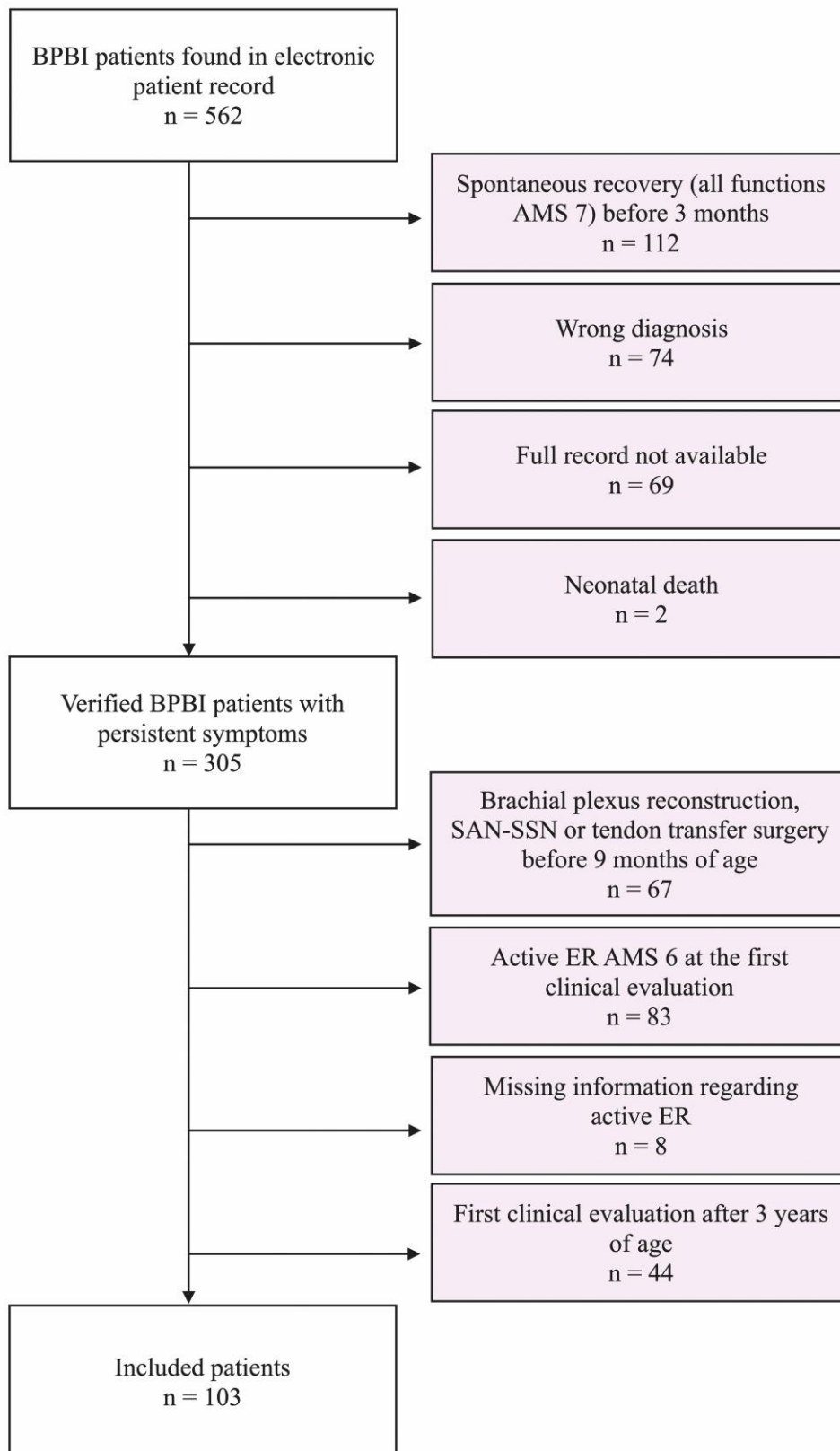


Figure 2

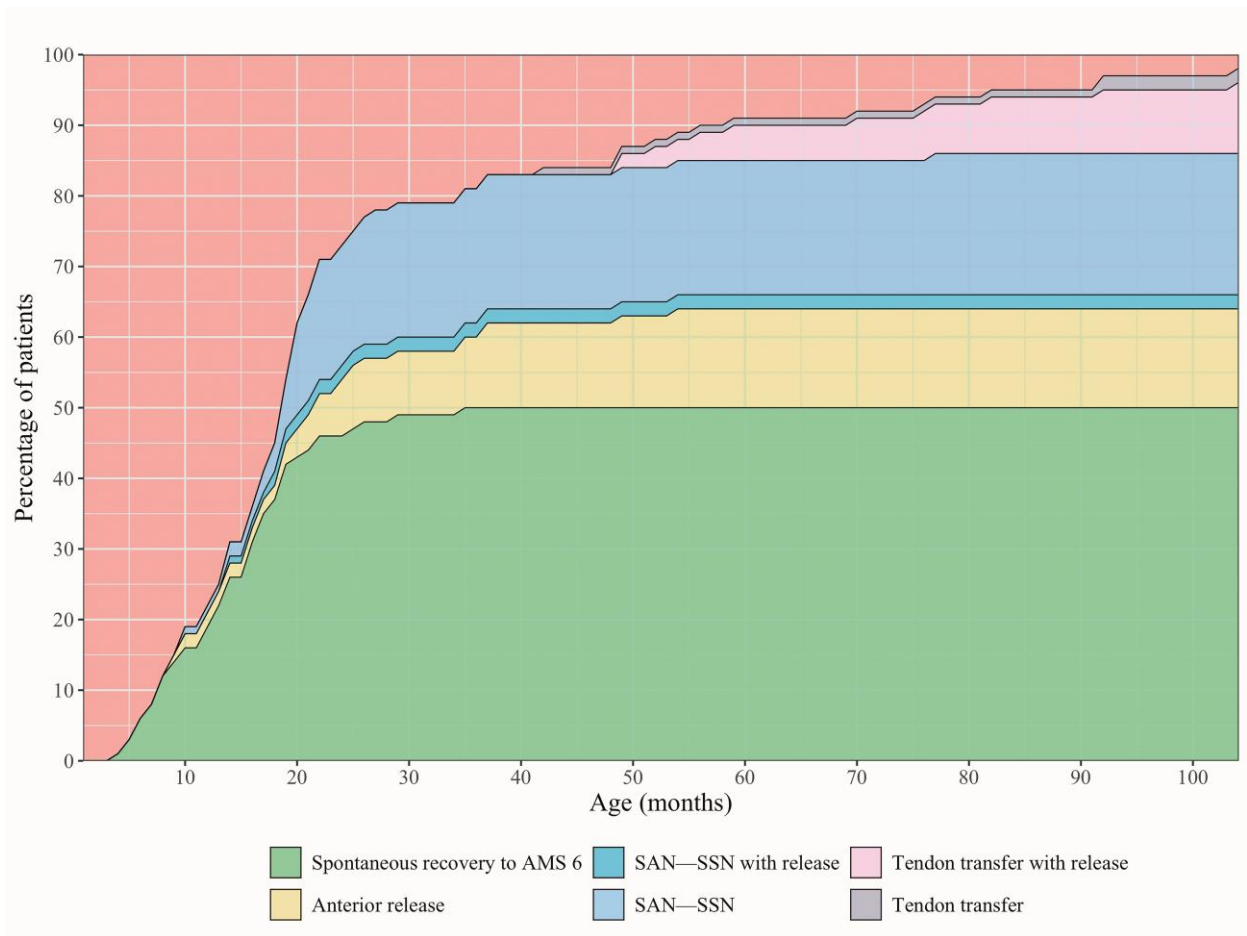


Figure 3

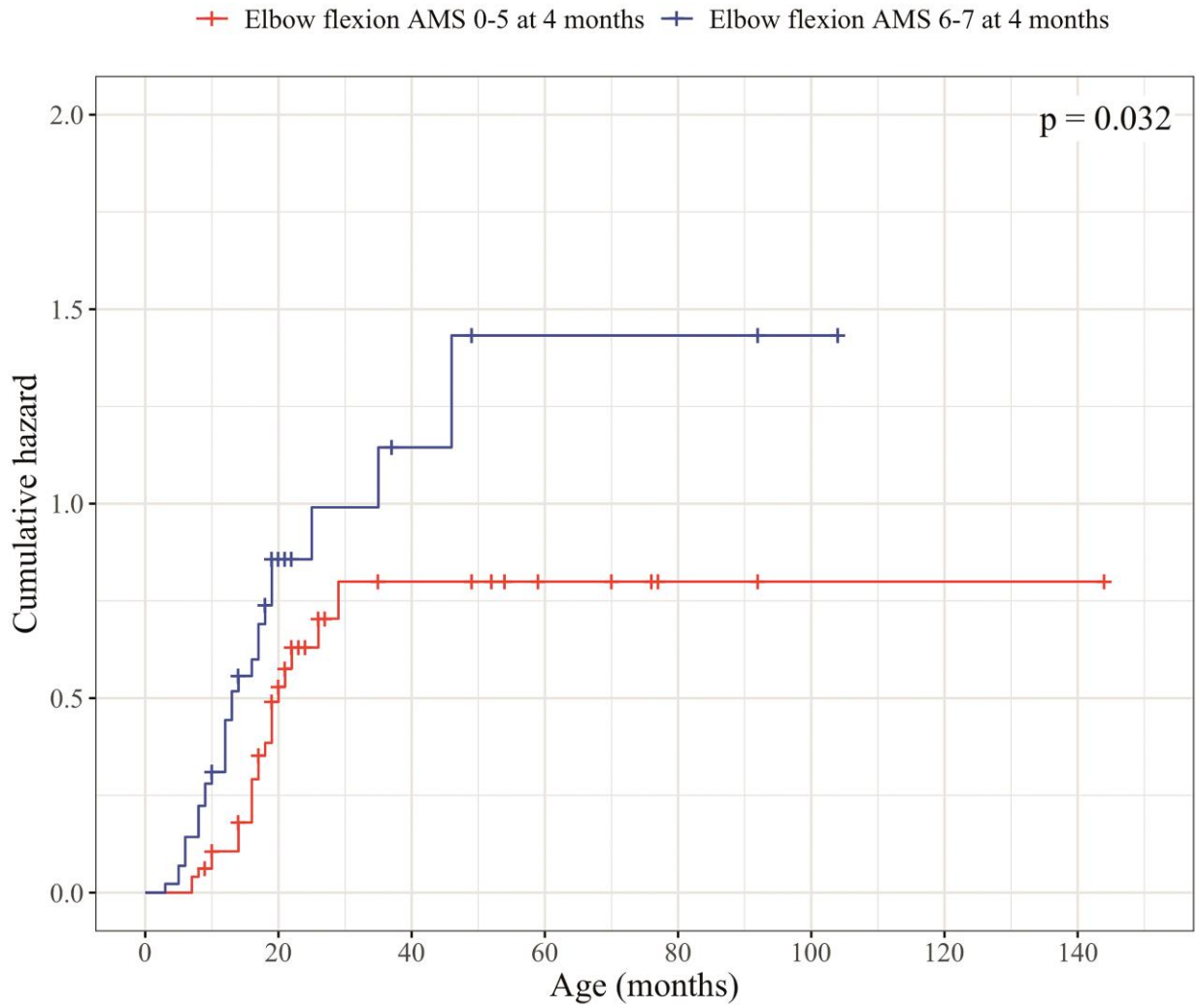


Figure 4

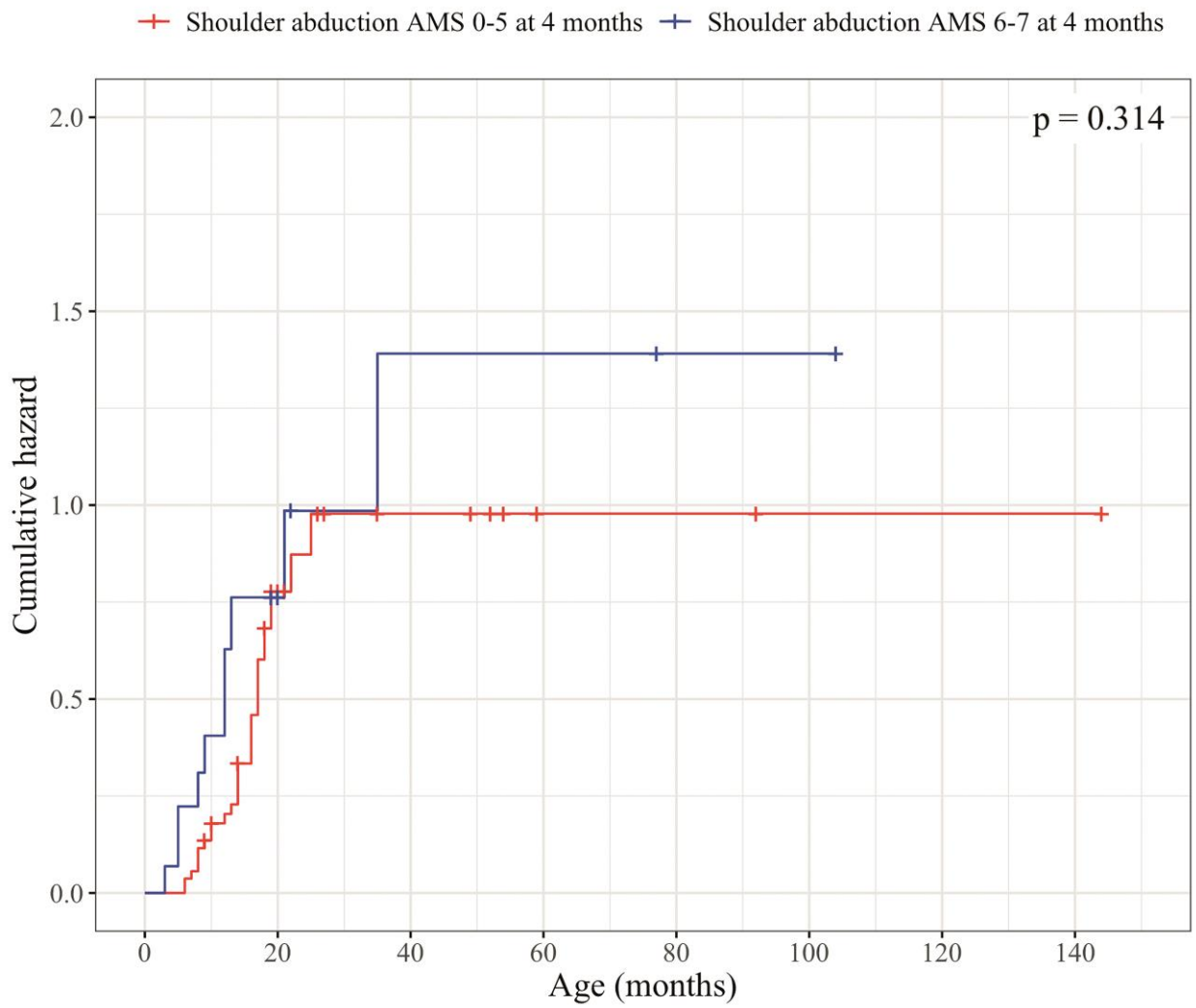
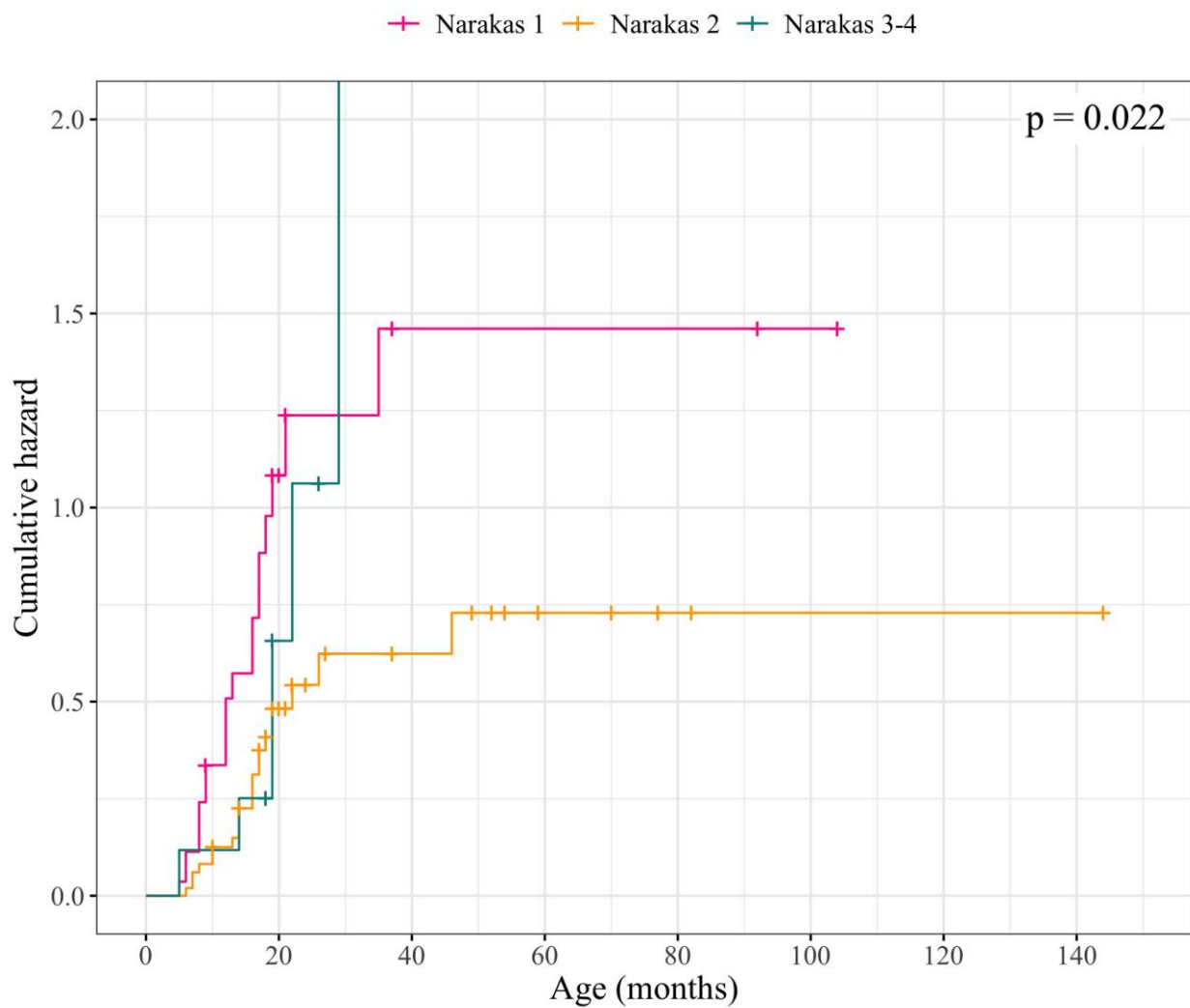


Figure 5



Table, Supplemental Digital Content 1. Cox proportional hazards regression for spontaneous recovery of shoulder external rotation (ER).

		CHR	95% CI	<i>p</i>-value	AHR	95% CI	<i>p</i>-value
Elbow flexion at 4 months				0.036*			0.035*
	AMS 0–5	1	–	–	1	–	–
	AMS 6–7	1.81	1.04–3.14	0.036*	1.69	0.79–3.61	0.178
Shoulder abduction at 4 months				0.349			0.558
	AMS 0–5	1	–	–	1	–	–
	AMS 6–7	1.42	0.69–2.94	0.335	0.61	0.25–1.46	0.263
Narakas at 1 month				0.029*			0.100
	Narakas 1	1	–	–	1	–	–
	Narakas 2	0.43	0.23–0.80	0.007*	0.43	0.20–0.93	0.032*
	Narakas 3 or 4	0.71	0.28–1.78	0.463	0.48	0.13–1.78	0.271

AMS = Active Movement Scale; CHR = crude hazard ratio; CI = confidence interval; AHR = adjusted hazard ratio