Shoulder internal rotation contracture formation in surgically managed C5, C6 brachial plexus birth injuries neurotmetic lesions fare worse than avulsions

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Shoulder Internal Rotation Contracture Formation in Surgically Managed C5, C6 Brachial Plexus Birth Injuries

Neurotmetic Lesions Fare Worse Than Avulsions

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Background: A typical feature in infants with severe C5-C6 brachial plexus birth injury (BPBI) requiring nerve repair is the formation of shoulder internal rotation contracture (IRC). The underlying pathophysiological mechanism is unknown, and the sequelae can be difficult to treat. The severity of the IRC differs among children. C5-C6 lesions are heterogeneous at the root level. Our null hypothesis was that the type of root-level lesion (axonotmesis or neurotmesis versus avulsion) was not associated with the extent of IRC formation over time in children with upper-trunk BPBI.

Methods: We performed a retrospective analysis of all patients with upper-trunk BPBI who underwent primary surgery of the C5 and/or C6 spinal nerves between 1990 and 2020 and had follow-up of at least 2 years. The primary outcome was passive shoulder external rotation (ER) in adduction at 1, 3, 5, 7, and 15 years of age. The secondary outcome was whether additional shoulder surgery was performed. The relationship between the nature of the C5-C6 lesion and IRC formation was analyzed using linear mixed models. The Kaplan-Meier method was used to estimate the cumulative risk of secondary shoulder procedures.

Results: In total, 322 patients were analyzed; mean follow-up was 7.2 ± 4.6 years. The C5-C6 root lesion type was significantly related to the passive range of ER (overall test in linear mixed model, p = 0.007). Children with avulsion of C5 and C6 (n = 21) had, on average, 18° (95% confidence interval [CI], 6.3° to 30°) less IRC formation than those with neurotmesis of C5 and C6 (n = 175) and 17° (2.9° to 31°) less than those with neurotmesis of C5 and avulsion of C6 (n = 34). IRC formation did not differ between the neurotmesis C5-C6 and neurotmesis C5-avulsion C6 groups. Secondary shoulder procedures were performed in 77 patients (10-year risk, 28% [95%CI, 23% to 34%]).

Conclusions: Shoulder IRC formation in infants with BPBI with surgically treated C5-C6 lesions occurs to a lesser degree if the C5 root is avulsed than when C5 is neurotmetic. This finding provides insight into the possible causative pathoanatomy and may ultimately lead to strategies to mitigate IRC.

Level of Evidence: Prognostic Level III. See Instructions for Authors for a complete description of levels of evidence.

One of the major therapeutical challenges in the treatment of children with C5-C6 brachial plexus birth injury (BPBI) is the formation of shoulder internal rotation contracture (IRC). IRC reduces the ability to position the hand in space, and thereby impairs function and quality of life[1-3]. Approximately 50% of all patients with BPBI develop IRC that does not resolve spontaneously[4]. Longstanding IRC can cause glenohumeral incongruence due to osseous deformities and dysplastic changes in the developing shoulder. The prevalence of glenohumeral deformities in children with BPBI is 33%[4]. IRC is the most common reason for secondary surgery following BPBI[5]. Opinions are divided on whether these osseous deformities are reversible after surgical intervention during growth and on the appropriate timing of the surgical intervention[6-9]. A clear understanding of the etiology by which BPBI causes IRC is lacking. A better

Disclosure: The **Disclosure of Potential Conflicts of Interest** forms are provided with the online version of the article (http://links.lww.com/JBJS/H232).

understanding of the underlying pathophysiology would enhance treatment possibilities.

The type of C5-C6 spinal root lesion in BPBI varies from axonotmesis to neurotmesis or avulsion[10]. It was anecdotally mentioned that patients with BPBI with avulsion injury show less IRC formation[11]. However, this observation has never been systematically analyzed, to our knowledge.

The aim of the current study was to analyze the relationship between IRC formation (clinically measured as the passive range of external rotation [ER] in adduction) and the type of C5-C6 nerve lesion, to identify factors that contribute to this so far elusive and difficult-to-treat phenomenon. Our null hypothesis was that the type of root-level lesion (axonotmesis or neurotmesis versus avulsion) was not associated with the extent of IRC formation over time in children with upper-trunk BPBI.

Materials and Methods

Population

From 1990 to 2020, a total of 1,817 infants with BPBI were referred to the Leiden University Medical Center, a multidisciplinary tertiary referral center for nerve injuries in the Netherlands. All consecutive patients with an upper BPBI who underwent primary brachial plexus (BP) exploration of the C5 and/or C6 spinal nerve(s) with at least 2 years of follow-up were included. An upper BPBI was defined as a C5-C6 BPBI with or without partial C7 involvement. Patients were selected on the basis of their clinical presentation. A C5-C6 lesion was defined as paralysis or severe weakness (Medical Research Council [MRC] scale[12] grade of <3) of the shoulder muscles and elbow flexors. Patients with an additional partial C7 lesion presenting with wrist-extensor weakness (MRC 2 to 4) were included. Infants without wrist-extension function (MRC 0 to 1) were excluded, regardless of the status of the C7 nerve on magnetic resonance imaging (MRI). Infants with diminished hand function indicative of C8-T1 involvement, and those who underwent reconstruction of the C7 root, were excluded as well.

We performed a retrospective analysis of postoperative passive range of shoulder ER in adduction. The follow-up period ended at the date of undergoing secondary surgery (tendon transfer of the latissimus dorsi or teres major to the posterolateral humeral head and/or an anterior release [opening of the rotator-cuff interval and transection of the coracohumeral ligament] or, in the early years, a sliding subscapularis release).

Patient Evaluation

If, at 3 months of age, elbow flexion with supination was absent in the supine position with the arm in 90° of abduction, computed tomographic (CT) myelography[13] (before 2015) or MRI[14] (in 2015 or later) to detect root avulsions and ultrasound of diaphragmatic excursion were performed. The BP was explored at 4 to 5 months if spontaneous recovery of anti-gravity elbow flexion did not occur or earlier if root avulsions were detected. Our surgical procedure has been described in detail elsewhere[15]. In short, the diagnosis of the lesion type for each root (i.e., axonotmesis, neurotmesis, or root avulsion) was based on evaluation of the extent and the location of neuroma formation, direct nerve stimulation, inspection of the spinal nerve at the intraforaminal level, imaging, and intraoperative histology. The type of injury was judged as axonotmetic when there was only mild neuroma formation and forceful contraction of related muscles on direct stimulation after external neurolysis. Neurotmetic lesions showed abundant neuroma formation, while no or only mild contraction could be felt on direct electrical stimulation. The suitability of the proximal nerve stump to serve as the axon source for grafts was verified using intraoperative frozen section examination. A nerve was considered avulsed when there was no neuroma formation and muscle contractions to root stimulation could not be elicited.

We used IRC assessment at the ages closest to 1, 3, 5, 7, and 15 years. These follow-up time points were standardized in the international PLexus oUtcome sTudy grOup (iPLUTO) consensus paper[16]. As follow-up time points had not yet been standardized for this cohort, we chose time points as close as possible to these ages. The passive range of ER in adduction, which reflects the degree of IRC, was assessed with the arm to the side with a 90° flexed elbow, starting from complete internal rotation and ending with the first sign of resistance during passive ER of the arm (Fig. 1)[17]. This movement was repeated several times to assess the angle of fixed contracture. Examinations were performed by our multidisciplinary team, which consists of attending surgeons, a physiatrist, a pediatric physical therapist, and an occupational therapist.

Statistical Analysis

SPSS (version 25; IBM) was used for statistical analysis. The association between the surgical diagnosis of the C5 and C6 spinal nerves and the passive range of ER in adduction was analyzed using linear mixed models to account for repeated measures. The linear mixed model included a random intercept and a scaled identity covariance matrix. The relation of other factors (sex, side of lesion, presentation, instrumented delivery, birth weight, and age at surgery) with IRC formation was also

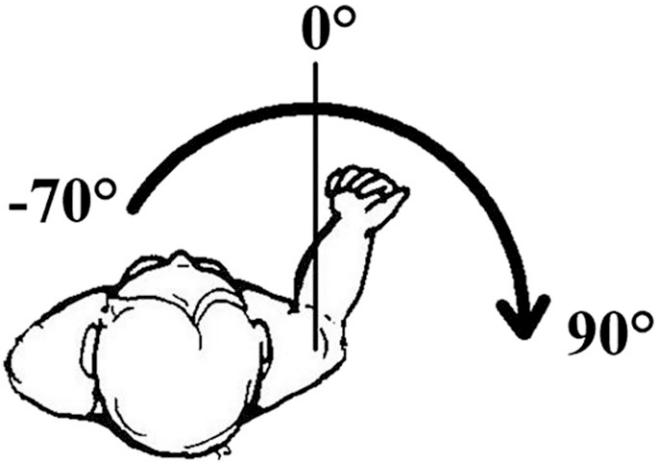

Fig. 1:
Drawing showing the evaluation of the passive range of external rotation in adduction, which was defined as the angle between the sagittal plane (0°) and the position of the 90° flexed elbow after passive external rotation in adduction.

analyzed. The Kaplan-Meier method was used to estimate the cumulative risk of subsequent shoulder procedures, which was the secondary outcome, and the log-rank (Mantel-Cox) test was used to compare survival distributions. A p value of <0.05 was considered significant.

No formal sample-size calculation was performed. We analyzed all data as available from the cohort of infants with BPBI referred to our center. In terms of effect sizes, 2 groups of 63 patients each would give 80% power to detect a medium effect size of 0.5. With repeated measurements, the statistical power is larger than with a single measurement. With a sample size of >300 patients, the statistical power was sufficient to address our research questions. The degree of certainty in the results is reflected with 95% confidence intervals (CIs).

Source of Funding
No external funding was received for any aspect of this work.

Results

In total, 322 patients met the inclusion criteria (Table I, Fig. 2). The uncensored follow-up for 11 of the included patients was <2 years because they developed IRC early and underwent early shoulder surgery; 66 had shoulder surgery after the age of 2 years. The median age at BP surgery was 5.0 months (interquartile range [IQR], 4.0 to 6.0 months). The type of lesion at the root level was axonotmesis of C5 and C6 in 38 patients (12%), neurotmesis of C5 and C6 in 175 patients (54%), neurotmesis of C5 and avulsion of C6 in 34 patients (11%), avulsion of C5 and C6 in 21 patients (7%), and mixed or partial C5-C6 lesions in 54 patients (17%) (Tables I and II). A total of 322 children were assessed at age 1 (0.5 to 1.9 years; mean [and standard deviation], 1.0 ± 0.1 years), 302 at age 3 (2.0 to 4.4 years; mean, 2.9 ± 0.4 years), 182 at age 5 (4.5 to 6.4 years; mean, 5.2 ± 0.5 years), 153 at age 7 (6.5 to 12 years; mean, 7.5 ± 1.0 years), and 81 at age 15 (12 to 19 years; mean, 14 ± 1.4 years). Some assessments were incomplete or censored; the number of assessments is given in Table III.

The passive range of ER in adduction was significantly associated with the C5-C6 root lesion type (overall test in linear mixed model, p = 0.007) and age (p < 0.001). IRC differences were present from 1 year of age onward (Table III). Differences were most pronounced for the avulsion C5-C6 group, which had, on average, an IRC that was 18° (95% CI, 6.3° to 30°) less than the IRC that of the neurotmesis C5-C6 group (Fig. 3-A). The neurotmesis C5-C6 and neurotmesis C5-avulsion C6 groups did not differ (Fig. 3-B). The avulsion C5-C6 group showed, on average, 17° (95% CI, 2.9 to 31°) less contracture formation than in the neurotmesis C5-avulsion C6 group (Fig. 3-C). There were no differences in the extent of IRC between the C5-C6 and C5-C6(C7) lesion types. There was no significant interaction between surgical diagnosis and age: in all diagnostic groups, the IRC increased at a similar rate in time after age 1. With an increase in age of 1 year, the passive range of motion decreased, on average, 2.6° (95% CI, 2.2° to 3.0°; p < 0.001). Other factors (sex, side of lesion, presentation, instrumented delivery, birth weight, and age at surgery) were nonsignificant.

TABLE I Patient Characteristics*

No.	322
Sex	
Male	142 (44%)
Female	180 (56%)
Affected side	
Left	150 (47%)
Right	167 (52%)
Bilateral	5 (1.6%)
Presentation at birth (n = 320)	
Cephalic	289 (90%)
Breech	31 (10%)
Instrumented delivery (n = 302)†	130 (40%)
Birth weight (n = 317)	
Mean ± SD *(kg)*	4.1 ± 0.7
Neurological deficit	
C5, C6	177 (55%)
C5, C6 (C7)	145 (45%)
Age at first outpatient visit	
Median (IQR) *(mo)*	2.7 (1.6-3.4)
Age at the time of surgery	
Median (IQR) *(mo)*	5.0 (4.0-6.0)
Surgical diagnosis, upper trunk‡	
Axonotmesis C5 and C6	38 (12%)
Neurotmesis C5 and C6	175 (54%)
Neurotmesis C5 and avulsion C6	34 (11%)
Avulsion C5 and C6	21 (7%)
Mixed or partial lesions	54 (17%)
No. of root avulsions	
None	241 (75%)
1	51 (16%)
2	28 (9%)
3	2 (0.6%)
Follow-up *(yr)*	
Mean ± SD	7.2 ± 4.6
Range	0.5-19

*SD = standard deviation, and IQR = interquartile range. Unless stated otherwise, values are displayed as the count, with the percentage in parentheses. †Forceps or vacuum extraction. ‡Axonotmesis includes axonotmesis/partial neurotmesis. Neurotmesis includes neurotmesis/subtotal neurotmesis/intraforaminal neurotmesis. Avulsion includes avulsion/subtotal avulsion/probable avulsion. Mixed or partial lesions = all other combinations of lesions (axonotmesis/partial neurotmesis of C5 and neurotmesis [n = 16], partial avulsion [n = 1], or avulsion [n = 9] of C6; neurotmesis of C5 and axonotmesis [n = 14] or partial avulsion [n = 2] of C6; partial avulsion of C5 and axonotmesis [n = 1], partial avulsion [n = 2], or avulsion [n = 5] of C6; or avulsion of C5 and neurotmesis [n = 2] or partial avulsion [n = 2] of C6).

Of the 322 patients, 77 patients (10-year cumulative incidence, 29%) with an IRC underwent secondary shoulder surgery. The indications for secondary surgery were twofold,

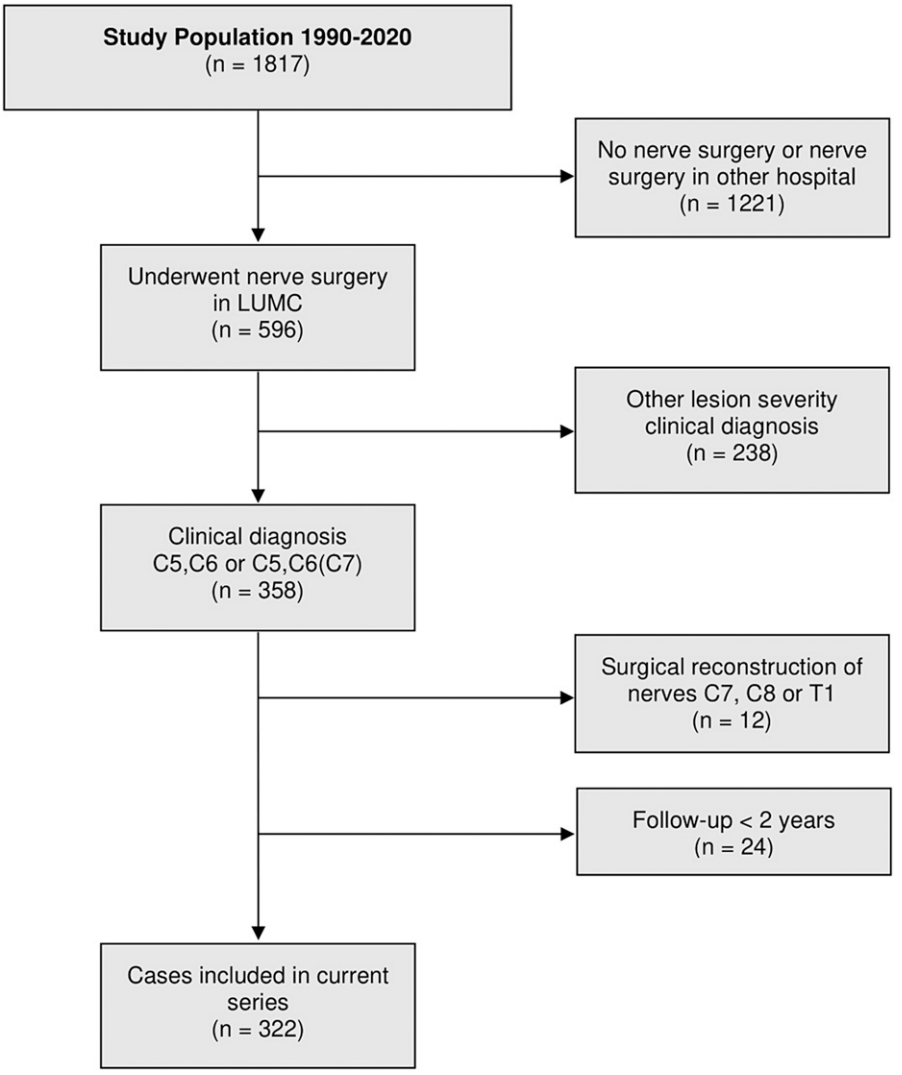

Fig. 2

Flowchart of the inclusion of patients in the current series. LUMC = Leiden University Medical Center.

TABLE II Surgical Diagnosis of the Upper Trunk in 322 Consecutive Surgical Cases, 1990 to 2020*

		C6 Diagnosis				
		Axonotmesis	Neurotmesis	Partial Avulsion	Avulsion	Total
C5 diagnosis	Axonotmesis	38 (12%)	16 (5.0%)	1 (0.3%)	9 (2.8%)	64 (20%)
	Neurotmesis	14 (4.3%)	175 (54%)	2 (0.6%)	34 (11%)	225 (70%)
	Partial avulsion	1 (0.3%)		2 (0.6%)	5 (1.6%)	8 (2.5%)
	Avulsion		2 (0.6%)	2 (0.6%)	21 (6.5%)	25 (7.8%)
	Total	53 (17%)	193 (60%)	7 (2.2%)	69 (21%)	322 (100%)

*Values are displayed as the number, with the percentage of the total cohort (n = 322) in parentheses. Axonotmesis includes axonotmesis/partial neurotmesis. Neurotmesis includes neurotmesis/subtotal neurotmesis/intraforaminal neurotmesis. Avulsion includes avulsion/subtotal avulsion/probable avulsion.

TABLE III Passive Range of Motion and Secondary Surgery in the Surgical Diagnosis Groups*

	AxC5-AxC6 (N = 38)	NC5-NC6 (N = 175)	NC5-AC6 (N = 34)	AC5-AC6 (N = 21)	Mixed or Partial (N = 54)
Passive ER†					
1 yr of age	47 ± 30 (n = 34)	46 ± 35 (n = 167)	47 ± 27 (n = 31)	57 ± 27 (n = 18)	56 ± 31 (n = 50)
3 yrs of age	54 ± 30 (n = 28)	43 ± 28 (n = 158)	47 ± 34 (n = 31)	59 ± 27 (n = 17)	55 ± 25 (n = 49)
5 yrs of age	32 ± 35 (n = 17)	34 ± 28 (n = 96)	35 ± 34 (n = 17)	49 ± 26 (n = 16)	45 ± 29 (n = 28)
7 yrs of age	26 ± 22 (n = 12)	32 ± 25 (n = 82)	36 ± 20 (n = 18)	48 ± 21 (n = 12)	35 ± 24 (n = 24)
15 yrs of age	23 ± 26 (n = 12)	26 ± 29 (n = 40)	23 ± 16 (n = 8)	38 ± 37 (n = 7)	17 ± 16 (n = 11)
Secondary surgery					
10-yr risk (95% CI)†	37% (18%-55%)	31% (23%-38%)	28% (11%-44%)	11% (−3.5%-26%)	21% (10%-36%)

*AxC5-AxC6 = axonotmesis of C5 and C6, NC5-NC6 = neurotmesis of C5 and C6, NC5-AC6 = neurotmesis of C5 and avulsion of C6, AC5-AC6 = avulsion of C5 and C6, and mixed or partial = mixed or partial upper-trunk lesions. †Passive ER = passive external rotation in adduction in degrees; values are displayed as the mean and the standard deviation (number of assessments). ‡CI = confidence interval.

either a progressive IRC of >30° and glenohumeral dysplasia of greater than grade 1 (in our cohort, these children were usually <5 years of age) or a functional lack of active ER (usually in children >5 years of age). Sixty-seven patients (cumulative incidence, 26%) had a tendon transfer of the latissimus dorsi or teres major to the posterolateral humeral head with or without an anterior or subscapularis release; 10 patients (cumulative incidence, 3.8%) underwent an anterior or subscapularis release without muscle transposition. The cumulative risk of a subsequent shoulder procedure in our population was 21% (95% CI, 16% to 26%) at age 5 and 28% (95% CI, 23% to 34%) at age 10. The 10-year cumulative risks per diagnosis groups are specified in Table III and Figure 4; log-rank analysis demonstrated that the differences between the groups did not reach significance.

Discussion

IRC is common in children with a C5-C6 BPBI lesion. The pathophysiology of IRC is not well understood, and treatment of severe IRC is challenging. In this study, we showed that, in patients who underwent BP exploration with or without nerve reconstruction, IRC was more severe if they had neurotmesis of C5 and C6 than if they had avulsion of C5 and C6. Because the neurotmesis C5-C6 and neurotmesis C5-avulsion C6 groups did not differ, but the avulsion C5-C6 and neurotmesis C5-avulsion C6 groups did, we conclude that the C5 lesion type, i.e., neurotmesis or avulsion, is associated with IRC formation. Previously, it was suggested that IRC was less severe in C5-C6 avulsion injury[11]. This was based on the observation that infants with BPBI born after a breech delivery showed less IRC. BP lesions following a breech delivery are associated with root avulsions, hence this indirect relationship was suggested[18,19]. Additionally, experimental studies comparing preganglionic and postganglionic C5-C6 injuries showed differences in contracture formation[20]. In our patients, IRCs were already present at 1 year of age. Therefore, the process underlying IRC formation must have taken place within the first year. Additionally, after 1 year, IRCs increased in all groups at a similar rate over time, suggesting that factors causing additional deterioration are similar for all groups. An increase over time did not occur in all children. Those without contracture formation and with good range of motion were discharged before 5 years of age.

Currently, 2 explanations regarding the cause of IRC formation prevail: (1) muscle imbalance between functioning internal rotators and paralyzed external rotators[5,21-25] and (2) impaired longitudinal growth of paralyzed muscles[26-28]. A study in mice demonstrated that denervation of the external rotators resulted in shoulder IRC, but surgical excision of the external rotators alone did not, suggesting that muscle imbalance cannot be solely responsible for contracture formation[28]. It was hypothesized that the preservation of afferent innervation of muscle spindles in preganglionic BPBI preserved muscle growth and protected against shoulder and elbow contractures[26-28]. Still, it is likely that some afferent neuronal cell bodies are preserved in neurotmesis, or at least in the case of a neuroma-in-continuity. Additionally, in surgically treated patients with BPBI, this preserving effect would be limited to the period before surgical reconstruction. During surgery for avulsion C5-C6 injury, the suprascapular nerve is cut to serve as an acceptor in a nerve transfer[29]. The preganglionic lesion of the C5 contribution to the suprascapular nerve is thereby turned into a postganglionic lesion, resulting in denervation of the muscle spindles after all. Only after axonal outgrowth, just as occurs after grafting a neurotmetic lesion, will reinnervation by afferent neurons of muscle spindles take place. The denervation after surgical neurotomy occurred, however, at around 5 months of age in the current cohort. Whether the inherent denervation of the muscle spindles at this age affects muscle development is unknown.

It is difficult to explain why the C5 lesion type is a notable factor. Details of motor innervation and the innervation of the shoulder joint by the C5 root may be relevant[30]. First, the C5 contribution to the long thoracic nerve remains intact in neurotmesis of C5, but not in avulsion of C5. Partial denervation of the upper part of the serratus anterior muscle with an avulsion of C5 might result in a scapular position that is less prone to IRC formation. We routinely stimulate the proximal C5 and C6 contributions to the long thoracic nerve during surgery, to differentiate between root avulsion and post-ganglionic lesions. Another detail might be that internal rotation by the pectoralis major

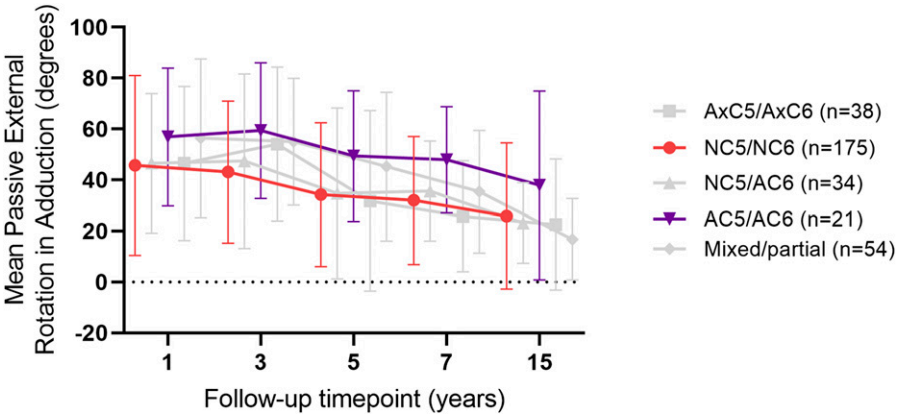

Fig. 3-A

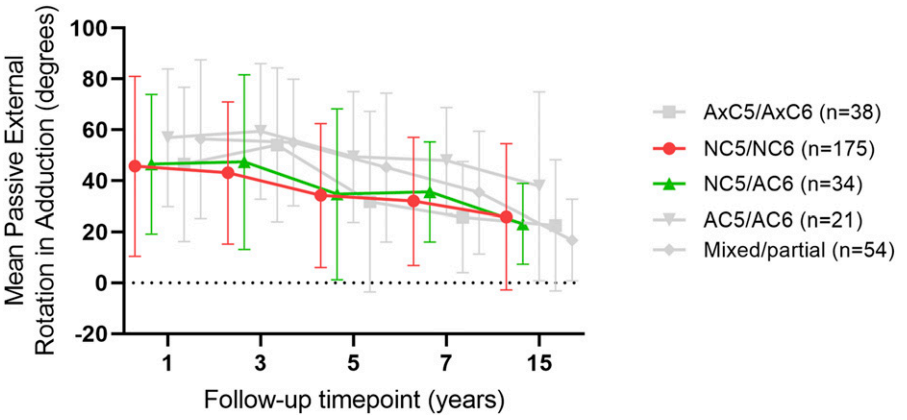

Fig. 3-B

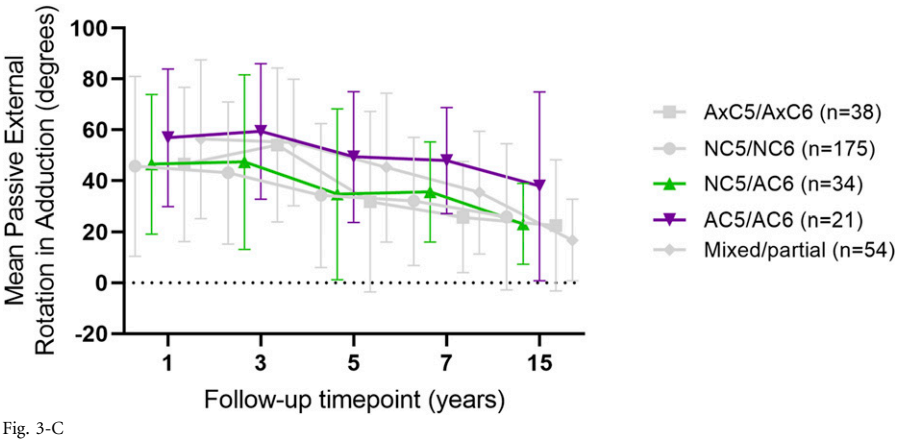

Fig. 3-C

Figs. 3-A, 3-B, and 3-C Group comparisons of passive external rotation (mean and standard deviation). **Fig. 3-A** Passive external rotation in the NC5/NC6 (red) and AC5/AC6 (purple) groups. **Fig. 3-B** Passive external rotation in the NC5/NC6 (red) and NC5/AC6 (green) groups. **Fig. 3-C** Passive external rotation in the NC5/AC6 (green) and AC5/AC6 (purple) groups . AxC5/AxC6 = axonotmesis of C5 and C6, NC5/NC6 = neurotmesis of C5 and C6, AC5/AC6 = avulsion of C5 and C6, and NC5/AC6 = neurotmesis of C5 and avulsion of C6.

muscles is more severely weakened with avulsion of C5 than with neurotmesis of C5. On the basis of our findings, we propose a third explanation for the underlying mechanism causing IRC. In neurotmetic C5-C6 lesions, there is some tension on the muscles, but they are rendered inactive by the misrouting of axons (possibly resulting in simultaneous contraction of the internal and external rotators), abnormal central programming, and impeded action-potential propagation[31,32]. In avulsion lesions, the muscles are completely flaccid. After birth, movements are made using "tricks," such as scapular instead of glenohumeral rotation. We hypothesize that the extent of IRC differs among lesion types at the root level because the trick movements differ between those with

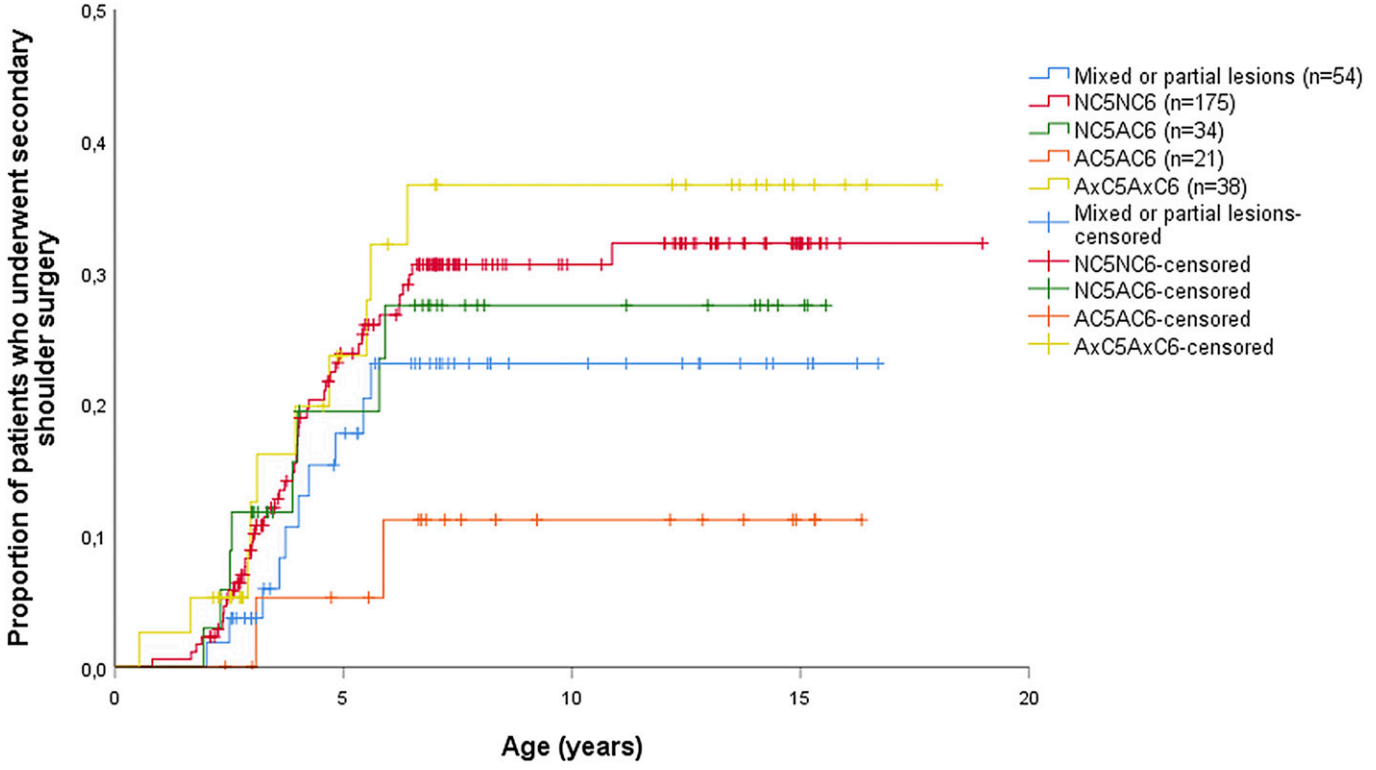

Fig. 4

Secondary shoulder surgery in the surgical diagnosis groups. NC5NC6 = neurotmesis of C5 and C6, NC5AC6 = neurotmesis of C5 and avulsion of C6, AC5AC6 = avulsion of C5 and C6, and AxC5AxC6 = axonotmesis of C5 and C6.

neurotmesis and the avulsion group. Clinically, IRC is associated with osseous deformities in the glenohumeral joint[25,33,34]. As contracture formation and glenohumeral deformities are intricately linked, it is plausible that both share causative factors[35]. There is some evidence that glenohumeral incongruence is greater with postganglionic BPBI, whereas preganglionic BPBI typically results in paralysis without substantial joint deformity[11,25,33,34,36]. In our study population, the 10-year risk of secondary procedures was 31% (95% CI, 23% to 38%) in the neurotmesis C5-C6 group compared with 11% (95% CI, −3.5% to 26%) in the avulsion C5-C6 group. Still, this finding was nonsignificant.

This study had limitations. First, it was subject to selection bias, as we only included patients who underwent BP surgery. Surgery was, however, the only way to verify the diagnosis of injury type at each root. We only selected children with upper-trunk lesions, to make the study population as homogeneous as possible. Although the resulting study population was quite large, the numbers in certain subgroups were relatively small. Furthermore, we excluded children with total C7 lesions. We based this diagnosis on the presence or absence of wrist-extension function, although it has been described that the extensor carpi radialis muscles are also innervated by C5 and C6[37]. As a result, it is impossible to completely rule out the potential effects of C7 spinal root denervation. Moreover, patients with good recovery were sometimes discharged early from follow-up; because of patient selection, outcomes reported in this study could be an underestimation of the actual degree of passive ER in adduction in our patients. Additionally, we excluded range-of-motion data from the analysis collected after patients underwent secondary shoulder surgery. This may have influenced outcomes because these are the patients with the most severe IRC. Finally, this study only reports on passive shoulder ER outcomes. Decisions about the management of BPBI should be made considering evidence regarding both sensory and motor outcomes of the entire upper limb. In this respect, the rate of secondary procedures may not be a reliable indicator of outcome, as the indication for secondary surgery may be based on a number of factors, which, besides the development of IRC, include active shoulder and elbow function, functional deficits of the child, indication shift over the years, and consent by the parents.

In conclusion, this large study of infants with surgically treated C5-C6 BPBI showed that IRC formation was less if both roots were avulsed than in neurotmetic C5-C6 lesions. Our null hypothesis could thus be rejected. More specifically, our study suggests that the lesion type of C5, neurotmetic or avulsion, is a factor that plays a role in IRC formation. This finding has wide-ranging implications for our understanding of IRC etiology and will guide improved prevention and treatment strategies for IRCs. ■

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