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Surgical Treatment of Congenital Radioulnar Synostosis in Children: Systematic Review

Yulia A. Fedorova, Sergei V. Vissarionov, Yaroslav N. Proschenko, Sofia A. Gevorgiz, Ekaterina A. Zakharyan

H. Turner National Medical Research Center for Children's Orthopedics and Trauma Surgery, St. Petersburg, Russia

Background. Congenital radioulnar synostosis (CRUS) may have a negative impact on the function of the upper limb and cause disability. The main aim of the surgical treatment is to correct the forearm position for diminishing functional limitations.

The study aimed to analyze the variety of surgical methods for correction of the pronation forearm deformity in children with CRUS based on the literature data.

Methods. We have searched publications in eLIBRARY, PubMed (MEDLINE), Ovid, ScienceDirect, Google Scholar databases. The analysis has included the age at surgery, indications for surgery, the target functional forearm position, the time of consolidation of the forearm bones, the frequency of neurovascular complications.

Results. Most authors considered subjective complaints as the main indication for surgical treatment, while some researchers recommended taking into account the forearm hyperpronation position. The median age of the surgical treatment was 5.17 years (3.25-9.46). The medians of the recommended forearm positions for unilateral CRUS were $0-10^{\circ}$ of pronation for the dominant, and $0-12.5^{\circ}$ of supination for the non-dominant limb; with bilateral cases $-0-17.5^{\circ}$ pronation for the dominant and $0-12^{\circ}$ supination for the non-dominant limb. Median of the osteotomy consolidation time varied from 6 to 8 weeks. The maximal time of forearm bone consolidation was significantly higher (p = 0.024) in the group with osteotomies through the synostosis site. Though the target forearm position was achieved in all cases, the number of complications in the proximal osteotomy group was statistically significantly different (p<0.01). The chances of neurovascular complications were 20.5 times higher in the group of patients who underwent osteotomy through the synostosis (95% CI: 2.7-155.6).

Conclusions. The problem of surgical treatment of children with CRUS in the world medical practice remains relevant despite the wide range of proposed methods. The development of an algorithm regarding the need for surgical treatment and its methodology requires further high-quality research.

Keywords: congenital radioulnar synostosis, derotation, rotation, forearm osteotomy, surgical treatment, child.

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Yulia A. Fedorova; e-mail: julsigareva@gmail.com

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Хирургическая коррекция положения предплечья у детей с врожденным радиоульнарным синостозом: систематический обзор литературы

Ю.А. Федорова, С.В. Виссарионов, Я.Н. Прощенко, С.А. Геворгиз, Е.А. Захарьян

ФГБУ «Национальный медицинский исследовательский центр детской травматологии и ортопедии им. Г.И. Турнера» Минздрава России, г. Санкт-Петербург, Россия

Актуальность. Врожденный радиоульнарный синостоз (ВРУС), будучи редкой аномалией развития, может оказывать существенное негативное влияние на функцию верхней конечности, затруднять самообслуживание. Основная задача хирургического лечения — коррекция положения предплечья с целью расширения функциональных возможностей.

Цель — анализ данных литературы о хирургических методах коррекции пронационной деформации предплечья у детей с ВРУС.

Материал и методы. Поиск публикаций выполнен в базах данных eLIBRARY, PubMed (MEDLINE), Ovid, ScienceDirect, Google Scholar. Проанализированы сроки консолидации костей предплечья, возраст хирургического лечения, показания к операции, целевое функциональное положение предплечья, частота нейроваскулярных осложнений в зависимости от варианта операции.

Результаты. Большинством авторов рекомендовано выполнение хирургического вмешательства при наличии субъективных жалоб, некоторые исследователи рекомендуют учитывать степень гиперпронационного положения предплечья. Медиана возраста хирургического лечения составила 5,17 лет (3,25-9,46). Медианы рекомендуемых целевых функциональных положений при одностороннем BPVC составили для доминантной конечности $0-10^\circ$ пронации, для субдоминантной $-0-12,5^\circ$ супинации; при двустороннем поражении $-0-17,5^\circ$ пронации для доминантной и $0-12^\circ$ супинации для субдоминантной конечностей. Медианы сроков консолидации зон остеотомии варьируют от 6 до 8 нед. Максимальные сроки консолидации костей предплечья в группе остеотомий через зону синостоза статистически значимо (p=0,024) выше в сравнении с группой остеотомий обеих костей предплечья. Несмотря на то, что целевое положение предплечья достигнуто во всех случаях, количество осложнений в группе проксимальных остеотомий статистически значимо отличалось (p<0,01). Шансы развития нейроваскулярных осложнений в 20,5 раз выше в группе пациентов, которым выполняли остеотомию через зону синостоза (95% ДИ: 2,7-155,6).

Заключение. Проблема хирургического лечения детей с ВРУС в мировой медицинской практике, несмотря на широкий спектр предложенных методик, остается актуальной. Разработка алгоритма определения необходимости хирургического лечения и его методики требует дальнейшего проведения исследований высокого качества.

Ключевые слова: врожденный радиоульнарный синостоз, врожденный лучелоктевой синостоз, остеотомия костей предплечья, хирургическое лечение, дети.



✓ Федорова Юлия Андреевна; e-mail: julsigareva@gmail.com

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BACKGROUND

Congenital radioulnar synostosis (CRUS) is a rare anomaly of the development of the upper limb that occurs in the early stages of embryogenesis as a result of a violation of differentiation from the common perichondria of the proximal parts of the radius and ulna [1]. Despite the fact that a single clinical case of idiopathic distal radioulnarl synostosis has been described in the literature [2], the authors suggest that the etiology of this condition is different.

The frequency of occurrence of CRUS in some regions of the European part of Russia is 0.47-1.29 per 100,000 population [3]. The world statistics of this disease are unknown.

Despite the relatively low frequency of occurrence, this anomaly has a significant impact on the function of the upper limb, especially in cases of severe pronation deformity and bilateral lesion. The existing congenital pathology significantly complicates the child's self-care: eating, holding objects and hygiene procedures, which is due to the absence or sharp restriction of the possibility of positioning the brush in the supination position. It should be noted that this pathology manifests and becomes pronounced with the acquisition of complex manual skills by the child [4]. These limitations become most obvious from the age of three [1], however, the anomaly may remain unnoticed until adolescence or even in an adult, especially with a unilateral lesion, a small synostosis and a forearm position close to the average physiological [5].

There are a number of studies in the modern literature devoted to attempts to restore active rotational movements in patients with congenital radioulnar synostosis [6-10]. The main task as a result of surgical treatment remains the correction of the forearm position in order to expand the functionality of the upper limb.

To date, more than 20 different options have been described for surgical correction of the hyperpronation position of the forearm in children with CRUS, but questions about the optimal age of surgery, indications for it, the most optimal and safe variant of derotation osteotomy, as well as alternative surgical treatment options aimed at restoring active rotational movements of the forearm are still being discussed.

The aim of this study was to analyze the literature data on surgical methods of correction of

pronational deformity of the forearm in children with congenital radioulnar synostosis.

METHODS

Search and selection of publications

This systematic review was carried out in accordance with the international requirements of PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses). The search for literary sources was carried out independently by two researchers (F.Y.A. and G.S.A.) in the electronic databases eLIBRARY, PubMed (MEDLINE), Ovid, ScienceDirect, Google Scholar using combinations of OR, AND operators and keywords for English-language papers "congenital radioulnar synostosis", "derotation", "rotation", "osteotomy", "surgical treatment", "children", "congenital radioulnar synostosis", "osteotomy", "children". The search query in the PubMed (MEDLINE) database included the following keywords: (congenital AND radioulnar AND synostosis) AND (surgical treatment OR derotation OR osteotomy) AND (child) NOT (trauma). Retrospectively, the search was not restricted, the date of the last request was January 31, 2022.

The criteria for inclusion:

- 1. A series of cases with more than 3 patients.
- 2. The age of patients at the time of surgical treatment is less than 18 years.
- 3. The use of surgical techniques for correcting the hyperpronation of the forearm in children with CRUS.

Due to the small number of analytical studies and the predominance of descriptions of clinical case report, studies with incomplete data presentation were included for analysis.

RESULTS

The initial search included 365 sources. After excluding duplicate papers, conference abstracts, book chapters, and comments, 283 studies were selected for screening. After analyzing the headings and abstracts of the articles, checking for compliance with the inclusion criteria, 26 articles directly met the goal of the study. The design of the articles was a description of a series of clinical observations, with the exception of the cohort study of Hwang et al. (2015) [11]. The research selection process is described in more detail in Figure 1.

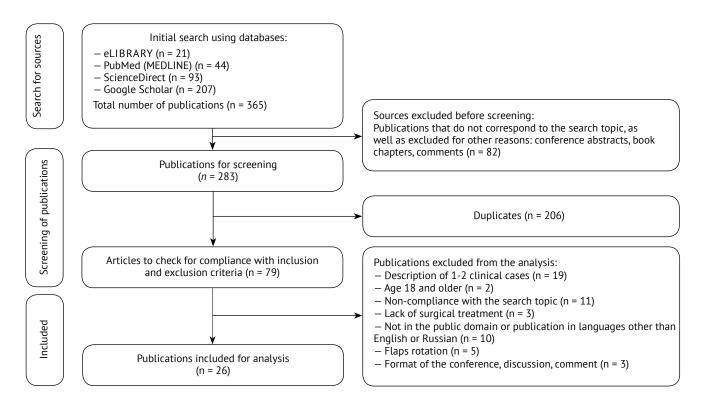
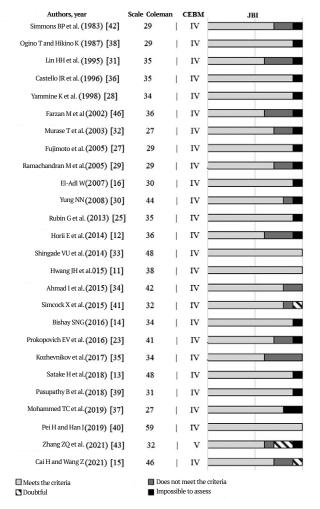


Fig. 1. Flowchart of the study



Risk of systematic error

The methodological quality of the selected studies was assessed in accordance with the criteria of the CEBM (Oxford Center for Evidence-Based Medicine) to determine the level of research, the JBI (Joanna Briggs Institute Critical Appraisal tools) and the Modified Coleman Scale (Modified Coleman Methodology Score) were used to assess the quality of the description of a series of clinical cases. Due to the fact that the overwhelming number of studies is a description of clinical cases series, an assessment on the Newcastle-Ottaw scale was not carried out. The results of the assessment are presented in Fig. 2.

Statistical analysis

The analysis of the extracted quantitative data (age at the time of surgery, the period of bone consolidation in the osteotomy area, the target functional position of the forearm) was performed using descriptive statistics methods after a preliminary check for the normality of the distribution according to the Shapiro-Wilk criterion.

Fig. 2. Results of the quality assessment of the included studies

To assess the frequency of neurovascular complications, the analysis of four-field tables was used. The comparison of consolidation periods in different osteotomy variants was performed using the Mann-Whitney U-test. Statistical processing was carried out in the IBM SPSS Statistics 26 program.

RESULTS AND DISCUSSION

Surgical options

Methods of surgical treatment of patients with congenital radioulnar synostosis can be divided into two main groups:

- surgery aimed at giving the forearm a functionally advantageous position (variants of derotation osteotomies) [12, 13, 14, 15, 16];
- surgery, the purpose of which was an attempt to improve rotational movements by resection of the synostosis zone with or without interposition of biological or synthetic materials [7, 8, 17, 18].

Historically, surgical separation of the synostosis zone and reconstructive techniques with an attempt to restore rotational movements seemed to be an ideal treatment option, but the final results according to most studies were not satisfactory.

Already in 1912, Dawson asked the question: "will it be possible to restore rotational movements when dividing the bone fusion zone?" [19]. A relapse of synostosis within 18 months after separation attempts was noted by a number of researchers [20, 21, 22, 23]. In 1998, Kanaya et al. [8] suggested filling the resection zone of synostosis with a blood-supplied fat graft. In this study, during 3.7 years of follow-up, none of the 7 patients relapsed, and the rotational function was preserved. In 2016, the 10-year long-term results of this technique were analyzed [9], which revealed a decrease in the amplitude of rotational movements, to a greater extent supination, the rate of extinction of rotation was about 16° per year. Sakamoto et al. in 2013 analyzed the results of treatment of 14 patients using the Kanaya method with an average follow-up period of 58 months [10]. Despite the fact that there was no recurrence of synostosis, rotational movements tended to fade [10]. In 2020, Dong et al. was retrospectively investigated the effectiveness of placing a blood-supplied flap in the radius resection zone in a large sample (36 patients). The authors

claimed to improve the rotational function with the achievement of pronation of 30.1° (15-45°), supination – $22,6^{\circ}$ (10-40°) [6].

It should be kept in mind that more than half of the cases of CRUS belong to type III according to the Cleary-Omer classification, that is, they are accompanied by an arcuate deformation of the radius, hypoplasia of the head of the radius. Consequently, the restoration of the rotational function of the forearm may be hindered by the pathological form of bone structures. Sakamoto (2013) et al. The emphasis was placed on the fact that in their study, a smaller rotation amplitude was achieved in patients with a larger arcuate deformity of the radius [10].

Thus, despite the good immediate results of attempts to restore the rotational function of the forearm, either an insufficiently long observation period is described, or a gradual extinction of rotation is noted.

At the moment, the leading methods of surgical treatment of CRUS are precisely the variants of forearm derotation osteotomies to achieve functionally advantageous position [24]. Conditionally, they can be divided as follows:

- surgery with gradual correction of pronation deformation using external fixation devices [25, 26];
- surgery consisting in performing a single-level osteotomy of the radius [12, 13, 23, 27];
- surgery accompanied by osteotomy of both forearm bones at different levels [11, 14, 15, 28, 29, 30, 31, 32, 33];
- surgery involving osteotomy through the synostosis zone [34, 35, 36, 37, 38, 39, 40, 41, 42, 43].

Most authors recommend performing surgery only in the presence of subjective complaints of the patient about the restriction of daily activity [4, 11, 33, 43, 44]. Some researchers recommend taking into account the severity and magnitude of the hyperpronation position of the forearm [28, 42]. When choosing a patient's treatment tactics and indications for surgical treatment, it is necessary to remember that the need for complex manual skills and the development of fine motor skills increase with the age of the child. In young children, even severe pronational deformities may not cause difficulties in providing self-service functions. A comparison of indications for surgical treatment is presented in Table 1.

Table 1
Comparative characteristics of indications for surgical treatment

Name	Osteotomy level	Surgery indications	Target functional position of the forearm
Green WT and Mital MA (1979) [45]	Through the synostosis zone	No information	With bilateral CRUS With 30-45° pronation for the dominant limb, 20-35° supination for the subdominant. With a one-sided CRUS of 10-20° supination
Simmons BP et al. (1983) [42]	Through the synostosis zone Distal radius	Forearm position ≥60° pronation – absolute indication, 15-60° – relative indication in the presence of subjective complaints; ≤15° – does not cause functional limitations	With a one-sided PRUS of 15° pronation, it is impractical to position the hand in the supination position. With a bilateral CRUS of 10-20° pronation for dominant, neutral for subdominant or taking into account the wishes of the patient
Ogino T and Hikino K (1987) [38]	Through the synostosis zone	Functional limitations, the presence of subjective complaints. There is no indication of the degree of pronation position of the forearm in degrees	With unilateral CRUS 0-20 ° supination. With a bilateral CRUS, 0-20° pronation for the dominant limb and 0-20° supination for the subdominant
Lin HH et al. (1995) [31]	Proximal ulna, distal radius	Functional limitations, the presence of subjective complaints. Unilateral pronation deformity ≥60° of the subdominant limb	20-30° pronation for the dominant limb, 0-20° supination for the subdominant
Castello JR et al. (1996) [36]	Through the synostosis zone	The position of the forearm ≥60° pronation with the presence of complaints is an absolute indication, 30-60° is a relative indication in the presence of complaints	0-15° of pronation
Yammine K et al. (1998) [28]	Ulna and radius diaphysis	The position of the forearm >900 or the restriction of daily activity with bilateral CRUS without specifying the degree of pronation position of the forearm in degrees	20° of pronation
Farzan M et al. (2002) [46]	Proximal ulna	Hyper-pronation position of the forearm without specifying degrees and bilateral lesion with functional limitations. In the absence of difficulties in daily activity, surgical treatment is not indicated	15° of supination
Murase T et al. (2003) [32]	Proximal ulna, distal radius	Functional limitations, the presence of subjective complaints. There is no indication of the degree of pronation position of the forearm in degrees	0-30° pronation for dominant limb, 0° for subdominant
Fujimoto M et al. (2005) [27]	Radius diaphysis	No information	10° pronation for dominant limb, 0° for subdominant
Ramachandran M et al. (2005) [29]	Ulna diaphysis, distal radius	The presence of subjective complaints, difficulty in self- service and daily activity, taking into account the severity of pronation deformation without specifying the degree in degrees	10° of supination
El-Adl W (2007) [16]	Proximal ulna, distal radius	Functional limitations, the presence of subjective complaints. There is no indication of the degree of pronation position of the forearm in degrees	30° pronation for dominant, 20° supination for subdominant
Hung NN (2008) [30]	Distal ulna, proximal radius	The presence of subjective complaints, difficulty in self-service and daily activity	0-30° pronation for dominant, neutral for subdominant with 70-100% correction of the initial deformation
Rubin G et al. (2013) [25]	Through the synostosis zone	Bilateral lesion with forearm position ≥90° pronation	0-30° supination, and for the subdominant limb more supinated
Horii E et al. (2014) [12]	Radius diaphysis	Forearm position ≤30° pronation does not require surgical correction	Neutral without specifying degrees
Shingade VU et al. (2014) [33]	Proximal ulna, distal radius	The presence of subjective complaints, objective functional limitations on the Failla or Jebsen-Taylor scale, regardless of the severity of the hyperpronation position	20-30° of supination
Hwang JH et al. (2015) [11]	Proximal ulna, distal radius	The presence of subjective complaints, difficulty in self- service and daily activity	0-30° of supination
Ahmad I et al. (2015) [34]	Through the synostosis zone	Functional limitations, the presence of subjective complaints, deformity >60°, taking into account a single or bilateral lesion	10-20° of supination

Name	Osteotomy level	Surgery indications	Target functional position of the forearm
Simcock X et al. (2015) [41]	Through the synostosis zone	Forearm position >60° with significant limitation of daily activity	10-20° of pronation
Bishay SNG (2016) [14]	Proximal ulna, distal radius	No information	20-30° pronation for the dominant limb, 20° supination for the subdominant
Prokopovich E.V. et al. (2016) [23]	Proximal radius	Forearm position >60° pronation	0-10° of pronation
Kozhevnikov O.V. and Kralina S.E. (2017) [35]	Through the synostosis zone	Functional limitations without a detailed description	Average physiological without specifying degrees
Satake H et al. (2018) [13]	Radius diaphysis	Forearm position ≥60° pronation	A position that allows for 90 ° supination of the hand, taking into account compensatory rotation at the level of the wrist joint
Pasupathy B et al. (2018) [39]	Two levels through the synostosis zone	Bilateral lesion with severe hyperpronation without specifying degrees, in other cases, the decision on surgical treatment is made individually	15-25° of supination
Mohammed TC et al. (2019) [37]	Through the synostosis zone	Forearm position =600 pronation with unilateral lesion with the presence of complaints, bilateral CRUS regardless of the degree of pronation position in degrees	0-30° of pronation
Pei X and Han J (2019) [40]	Through the synostosis zone	Forearm position ≥55° pronation, ≤ 10 points on the Failla scale	10° of pronation - 20° of supination
Zhang ZQ et al. (2021) [43]	Through the synostosis zone	The presence of subjective complaints, difficulty in self- service and daily activity	No information
Cai H and Wang Z (2021) [15]	Proximal ulna, distal radius	Forearm position >60° pronation, <8 points on the ADL scale	20° pronation for dominant limb, neutral for subdominant without specifying degrees

Optimal age for surgical treatment

A consensus on the optimal age for surgery according to the literature has not been formed at the moment. It is recommended to start surgical treatment no earlier than the age of two [47]. The youngest age of the operation for a child with CRUS, according to literary, was 1.5 y.o. [35]. It was found that surgical treatment at the age of over 7 years is associated with a high risk and a higher incidence of neurovascular complications compared to patients who underwent surgery at an early age [24]. According to the results of the analysis of the extracted data, the median age of surgical treatment was 5.17 years (3.25-9.46) (Table 2).

Assessment of upper limb function

The analysis of the functional results of surgical treatment using objective scales was carried out only in 23% of the publications (6 out of 26 papers) included in the analysis. Objective scales for assessing the function of the upper limb have been used since 2005, among them

– ADL (activity of daily living) [13, 15, 27, 33], qDASH [13], Failla [33, 40], Jabson-Taylor [33], Liverpool Elbow Score [11]. In another 9 papers (35%), the authors either offer their own scale without its detailed description, or assess the subjective satisfaction of patients with the result of surgical treatment. The most complete assessment of the function of the upper extremities is described in the paper of Shingade et al. (2014) [33].

Terms of consolidation and the option of fixing

Various ways of fixing the bones of the forearm are proposed – from the absence of internal fixation [12, 16, 26, 27, 31, 33] prior to the application of the external fixation device [25], osteosynthesis with K-wires is most common [14, 23, 29, 30, 32, 34, 35, 36, 37, 39, 41, 43]. Additionally, in all cases, the limb was immobilized with a posterior plaster splint from the upper third of the shoulder to the metacarpophalangeal joints when the elbow joint was bent at an angle of 90°. Hwang et

al. (2015) compared the groups with internal fixation and the absence of such, without revealing statistically significant differences in the terms of consolidation, the angle of correction, the postoperative position of the forearm, the magnitude of the loss of correction after surgery and the function of the upper limb according to the Liverpool scale [11]. Median periods of consolidation of osteotomy zones vary from 6 to 8 weeks (Table 2). A comparative analysis of the minimum and average consolidation periods in the groups that underwent osteotomy through the synostosis zone and osteotomy of both forearm bones revealed no statistically significant differences (Table 3).

When comparing the terms of consolidation after different variants of osteotomies, statistically significant differences were found in the maximum terms of consolidation (p=0.024).

The maximum periods of consolidation in the osteotomy group through the synostosis zone were higher in comparison with the osteotomy group of both forearm bones. The differences in the minimum and average consolidation periods were not statistically significant.

Forearm position and safe degree of correction

The optimal position of the forearm remains an actively discussed issue. Hwang et al. (2015) indicate that the position of excessive supination may limit the daily activity of the child due to global computerization and widespread use of the keyboard [11]. Shingade et al. (2014) are of the opinion that the peculiarities of hygienic procedures of the inguinal region require almost complete supination [3]. Many groups of authors, mainly from Asian

Table 2 The results of the analysis of the extracted quantitative data from the articles

	Extrac	ted data	Min, Me [IQR]	Average, Me [IQR]	Max, Me [IQR]	
Age at t	he time of su	ırgery, full years	3,25 [2,20-4,00]	5,17 [4,74-6,90]	9,46 [8,25-13,00]	
Consoli	dation period	d, weeks	6,0 [5,5-7,0]	7,0 [6,0-8,0]	8,0 [9,0-14,0]	
the	One-sided	Dominant limb	0 [-20,0-12,5]	-	10,0 [-5,0-25,0]	
tion of tl degrees	CRUS	Subdominant limb	-12,5 [20,0-0,0]	-	0 [-10,0-10,0]	
posi arm,	Two- sided	Dominant limb	0 [-17,5-15,0]	-	17,5 [0,0-30,0]	
Target forea	CRUS	Subdominant limb	-12,5 [-20,0-0,0]	-	0 [-12,5-5,0]	

The supination position is represented by negative values.

Table 3 The results of a comparative analysis of the terms of consolidation in various variants of rotational osteotomies of the forearm bones

	Surger	y option	
Parameter, weeks	Osteotomy through the synostosis zone	Osteotomy of both forearm bones at different levels	p
	Me [IQR]	Me [IQR]	
Minimum consolidation period	8,0 [5,0-8,6]	6,0 [5,0-6,9]	0,381
Average consolidation period	9,4 [7,0-12,0]	6,95 [5,9-7,5]	0,142
Maximum consolidation period	12,0 [10,1-16,0]	7,95 [7,0-9,0]	0,024*

^{* -} differences in parameters are statistically significant (p<0.05)

countries, emphasize the need to take into account the socio-cultural environment – for example, eating with chopsticks requires holding the bowl in the supination position [13, 38, 40]. A number of researchers have indicated that a small supination can be well compensated by shoulder retraction [11, 29, 33], in contrast to the pronation [12]. Other groups of authors emphasize the need to position the subdominant limb in a more "supination" position [14, 27, 31, 32, 38, 42]. The results of the Pei study et al. (2019) demonstrated the highest score on the Failla functional scale in patients with a forearm position of 0-20° supination, both for dominant and subdominant limbs [40]. A detailed comparison of the target positions of the forearm after derotation osteotomy is presented in Table 2. The medians of the recommended target functional positions for unilateral CRUS were 0-10° pronation for the dominant limb, 0-12.5° supination for the subdominant limb; for bilateral lesions - 0-17.5° pronation for the dominant and 0-12° supination for the subdominant limb (Table 2).

Despite the fact that the target position of the forearm was achieved in all cases, the number of complications differed depending on the level of osteotomy – the highest frequency of neurovascular complications was associated with osteotomy through the synostosis zone (Table 4).

When comparing the frequency of neurovascular complications depending on the surgical intervention option (through the zone of synostosis and both forearm bones at different levels), statistically significant differences were obtained (p<0.01). The chances of developing neurovascular complications increased in the group of patients who underwent osteotomy through the synostosis zone by 20.5 times (95% CI: 2.7-155.6). There was an average strength relationship between the compared signs (V = 0.235).

Other variants of possible complications, as well as more detailed characteristics of the surgical options used are described in Table 5.

CONCLUSION

According to the literature data, the problem of surgical treatment of children with congenital radioulnar synostosis in world medical practice remains relevant due to the lack of a unified approach, decision-making algorithm and clear indications for surgery. Based on the information presented and analyzed by us, we can summarize:

- 1. Many authors recommend performing surgical treatment of children with radioulnar synostosis before starting school, at the age of 5 years (3-9 years).
- 2. Insufficient attention is paid to an objective assessment of the function of the upper extremities when determining indications for surgical treatment using point scales and questionnaires.
- 3. The target functional position of the forearm differs depending on the leading limb and in the case of a bilateral lesion, more supination is preferable for the subdominant limb.
- 4. The terms of consolidation of the forearm bones in various variants of osteotomies vary from 6 to 8 weeks. There were no statistically significant differences in the minimum and average terms of consolidation in the group of osteotomies through the zone of synostosis and osteotomies of both forearm bones. At the same time, the maximum consolidation periods differ statistically significantly (p=0.024), averaging 12 weeks after osteotomy through the synostosis zone and 7.95 weeks with osteotomy of both forearm bones.
- 5. Despite the fact that the target functional position of the forearm was achieved in all variants of surgery, osteotomies through the zone of synostosis, as well as proximal osteotomies of the radius are accompanied by a higher frequency of neurovascular complications.

Thus, the development of a decision-making algorithm regarding the need for surgical treatment and its methodology remains an unsolved task to the end and requires further research.

	Complications	Neurocirculatory 4/20 Infectious 1/20 Loss of correction 3/20	Neurocirculatory 2/11	No	N O	Loss of correction 3/6 Slow consolidation 1/6	Neurocirculatory 1/3	Loss of correction 1/4	No	Neurocirculatory 1/5 Angular deformation 1/5	Neurocirculatory 1/5 Angular deformation 1/5	Loss of correction 5/34	Neurocirculatory 2/4 Infectious 1/4	No	Slow consolidation 1/36
	Observation period, months	150 (12-312)	>24	69 (20-165)	96 (36-120)	228,8 (12-264)	40 (12-60)	45,8 (14-73)	21 (12-36)	29 (18-43)	26,4 (13-38)	64 (30-129)	99,6 (84-120)	60 (12-132)	54±13 (36-84)
	Correction angle, degrees	67 (25-90)	70 (30-110)	No information	In 1 case, a two- stage correction due to derotation by 100 degrees	66 (40-90)	No information	(08-09) 59	No information	78 (50-90)	58,6 (35-110)	No information	60 гр + 4 гр/мес	No information	No information
Comparative characteristics of rotational osteotomies	Forearm position after surgery, degrees	In pronation	4.2 of supination (10 of pronation - 20 of supination)	20 of pronation (30 of pronation - 20 of supination)	0-15 of pronation	19 of pronation (30 of supination - 45 of pronation)	15,3 of supination (10-15 of supination)	7,5 of pronation (0-20 of pronation)	2,6 of pronation (10 of pronation - 10 of supination)	10 of supination	25 (20-30) of pronation for dominant, 25 (25-30) supination for subdominant limb	6 of pronation for dominant, 10 of pronation for subdominant (0-30 of pronation)	15 of supination (0-30)	Neutral (except 2 cases)	27,2±4,1 (20-30) of supination
characteristics of r	Forearm position before surgery, degrees of pronation	82 (45-120)	65,8 (30-110)	81,7 (60-90)	80-100	85 (30-110)	113,3 (110-120)	78 (70-80)	75 (70-85)	68 (40-80)	76 (65-85)	82 for the dominant and 74 for the subdominant limb (65-85)	100 (90-110)	72 (40-100)	56,3 13,7 (30-86)
Comparative	Consolidation period, weeks	No information	No information	No information	No information	There is no indication of deadlines	No information	7,5 (6,9-7,9)	12,0-20,0	6,3 (6-9)	5,9 (5-7)	7,2 (6,6-7,4)	9,4 (8,6-10,1)	No information	ъ
)	Age at the time of surgery, full years	8 (2,5-17,5)	7 (4-13)	4,5 (2-14,33)	7,33 (4-9)	4-12	4,6 (3-6)	3,9 (2,2-5)	4,42 (3,92-4,92)	4,9 (3,5-8,25)	5,5 (3,75-8,25)	6,25 (3,75-9,92)	11 (9-13)	5,1 (2,5-8,75)	8,6 ± 3,7
	The fixation option	Steinmann pin + K-wires/ brackets	K-wires + pin	No fixation	K-wires	Plate / ex-fix	K-wires	K-wires	No fixation	K-wires	No fixation	K-wires	Ex-fix	No fixation	No fixation
	Osteotomy level	Through the synostosis zone. Distal radius	Through the synostosis zone	Proximal ulna, distal radius	Through the synostosis zone	Ulna and radius diaphysis	Proximal ulna	Proximal ulna, distal radius	Radius diaphysis	Ulna diaphysis, distal radius.	Proximal ulna, distal radius	Proximal radius, distal ulna	Through the synostosis zone	Radius diaphysis	Proximal ulna, distal radius
	Author	Simmons BP et al. (1983) [42]	Ogino T and Hikino K (1987) [38]	Lin HH et al. (1995) [31]	Castello JR et al. (1996) [36]	Yammine K et al. (1998) [28]	Farzan M et al. (2002) [46]	Murase T et al. (2003) [32]	Fujimoto M et al. (2005) [27]	Ramachandran M et al. (2005) [29]	El-Adl W (2007) [16]	Hung NN (2008) [30]	Rubin G et al. (2013) [25]	Horii E et al. (2014) [12]	Shingade VU et al. (2014) [33]

End of Table 4

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Author	Osteotomy level	The fixation option	Age at the time of surgery, full years	Consolidation period, weeks	Forearm position before surgery, degrees of pronation	Forearm position after surgery, degrees	Correction angle, degrees	Observation period, months	Complications
Hwang JH et al. (2015) [11]	Proximal ulna, distal radius	2 groups – with and without axial fixation with spokes	7 (4-16)	7 (6-8)	47 (30-65)	27 of supination (25-30)	74 (55-90)	33 (12-72)	No
Ahmad I et al. (2015) [34]	Through the synostosis zone	K-wires	5,16 (3-11)	7 (5-12)	68,75 (45-90)	14,58 of supination (10-20 of supination)	77,91 (45-100)	62,28 (48-132)	Neurocirculatory 3/12
Simcock X et al. (2015) [41]	Through the synostosis zone	K-wires	6,8 (3,0-18,8)	8	85 (60-100)	8 (0-30)	78 (40 - 95)	46 (6-148)	Neurocirculatory 3/26
Bishay SNG (2016) [14]	Proximal ulna, distal radius	K-wires	5,17 (4,83 - 6,42)	(8-9) (6-9)	70,7 (60–85)	No information	59,8 (30–90)	30,4 (24–36)	No
Prokopovich E.V. et al. (2016) [23]	Proximal radius	K-wires, 1 patient – Plate	No information	No information	No information	No information	No information	6-120	Neurocirculatory - 4 cases (8,9%) Fracture of the osteosynthesis zone - 2 cases
Kozhevnikov O.V. and Kralina S.E. (2017) [35]	Through the synostosis zone	K-wires	1,5-9	No information	90-90	No information	45-60	12	Neurocirculatory 1/6
Satake H et al. (2018) [13]	Radius diaphysis	No fixation	4,5-10,0	9	51,3 (30-90)	4 of supination (20 of pronation - 30 of supination)	55 (30-90)	163,2 (120-230,4)	No
Pasupathy B et al. (2018) [39]	Through the synostosis zone	K-wires	3,8 (2-9)	No information	72,25 (55-85)	22 of supination (12-35)	No information	26,4 (7-48)	Infectious 1/20
Mohammed TC et al. (2019) [37]	Through the synostosis zone	K-wires	5 (3-9)	12 (8-16)	62 (45-85)	0-30 of pronation	54 (30-75)	No information	Neurocirculatory 1/10
Pei X and Han J (2019) [40]	Through the synostosis zone	Plate	$4,87 \pm 3,06$ $(2-13)$	∞	62,92±7,11 (55-80)	$7,94 \pm 7,25$ of supination	$70,86 \pm 9,58$ (50-90)	55,19 ± 27,10 (24 - 123)	Neurocirculatory 4/31
Zhang ZQ et al. (2021) [43]	Through the synostosis zone	K-wires	5,2 (4,3-6,0)	ιn	98 (95-100)	No information	60 (fixed value)	No information	No
Cai H end Wang Z (2021) [15]	Proximal ulna, distal radius	No fixation	6,25 (4-9)	80	75,31 (45-90)	15 of pronation	No information	27,4 (24-36)	No

DISCLAIMERS

Author contribution

Fedorova Yu.A. — concept and design of the study; the collection and processing of material, evaluation and interpretation of the data and the preparation of the text.

Vissarionov S.V. — study design; interpretation of the data, preparation and editing of the text.

Proschenko Ya.N. — analysis and interpretation of the data; preparation and editing of the text.

Gevorgiz S.A. — collection and processing of material; preparation of the text.

Zakharyan E.A. — data analysis; editing of the text.

All authors have read and approved the final version of the manuscript of the article. All authors agree to bear responsibility for all aspects of the study to ensure proper consideration and resolution of all possible issues related to the correctness and reliability of any part of the work.

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REFERENCES

- 1. Bhatt C.R., Mehta C.D. Case report: Congenital radioulnar synostosis and its embryological correlation and functional assessment. *J Anatomical Society of India*. 2011;60(2):236-238. doi: 10.1016/S0003-2778(11)80035-3.
- 2. Shoham Y., Gurfinkel R., Sagi A. Idiopathic distal radioulnar synostosis. *J Plast Surg Hand Surg*. 2014;48(1):89-90. doi: 10.3109/2000656X.2012.754626.
- Zinchenko R.A., Ginter E.K. [Hereditary diseases in human populations]. In: [National guideline. Hereditary diseases]. Ed. by N.P. Bochkova, E.K. Ginter, V.P. Puzyrev. Moscow: GEOTAR-Media; 2012. p. 662-704. (In Russian).
- 4. Kepenek-Varol B., Hoşbay Z. Is short-term hand therapy effective in a child with congenital radioulnar synostosis? A case report. *J Hand Ther.* 2020;33(3):435-442. doi: 10.1016/j.jht.2019.03.009.
- 5. Alsharif M.H.K., Almasaad J.M., Taha K.M., Elamin A.Y., Bakhit N.M., Noureddin M.A. et al. Congenital radioulnar synostosis presenting in adulthood a case report. *Pan Afr Med J.* 2020;36:75. doi: 10.11604/pamj.2020.36.75.21413.

- 6. Dong Y., Hu P., Liu F., Niu X., Feng G., Wang F. [Treatment of congenital radioulnar synostosis with radial derotational osteotomy and local subcutaneous pedicled fat flap filling in fusion area]. *Zhongguo Xiu Fu Chong Jian Wai Ke Za Zhi*. 2020;34(7):820-825. (In Chinese). doi: 10.7507/1002-1892.201911079.
- 7. Funakoshi T., Kato H., Minami A., Suenaga N., Iwasaki N. The use of pedicled posterior interosseous fat graft for mobilization of congenital radioulnar synostosis: a case report. *J Shoulder Elbow Surg.* 2004;13(2):230-234. doi: 10.1016/j.jse.2003.09.009.
- Kanaya F., Ibaraki K. Mobilization of a congenital proximal radioulnar synostosis with use of a free vascularized fascio-fat graft. *J Bone Joint Surg Am.* 1998;80(8): 1186-1192. doi: 10.2106/00004623-199808000-00012.
- 9. Kanaya K., Iba K., Yamashita T. Long-term results after a free vascularized adipofascial graft for congenital proximal radioulnar synostosis with an average follow-up of 10 years: a series of four cases. *J Shoulder Elbow Surg.* 2016;25(8):1258-1267. doi: 10.1016/j.jse.2016.04.009.
- Sakamoto S., Doi K., Hattori Y., Dodakundi C., Montales T. Modified osteotomy (Kanaya's procedure) for congenital proximal radioulnar synostosis with posterior dislocation of radial head. *J Hand Surg Eur Vol.* 2014;39(5):541-548. doi: 10.1177/1753193413493386.
- 11. Hwang J.H., Kim H.W., Lee D.H., Chung J.H., Park H. One-stage rotational osteotomy for congenital radioulnar synostosis. *J Hand Surg Eur Vol.* 2015;40(8):855-861. doi: 10.1177/1753193415580066.
- 12. Horii E., Koh S., Hattori T., Otsuka J. Single osteotomy at the radial diaphysis for congenital radioulnar synostosis. *J Hand Surg Am.* 2014;39(8):1553-1557. doi: 10.1016/j.jhsa.2014.05.018.
- 13. Satake H., Kanauchi Y., Kashiwa H., Ishigaki D., Takahara M., Takagi M. Long-term results after simple rotational osteotomy of the radius shaft for congenital radioulnar synostosis. *J Shoulder Elbow Surg.* 2018;27(8):1373-1379. doi: 10.1016/j.ise.2018.04.012.
- 14. Bishay S.N. Minimally invasive single-session double-level rotational osteotomy of the forearm bones to correct fixed pronation deformity in congenital proximal radioulnar synostosis. *J Child Orthop.* 2016;10(4):295-300. doi: 10.1007/s11832-016-0750-8.
- 15. Cai H., Wang Z. Treatment of children's proximal radioulnar joint fusion through single dual-plane osteotomy on combination with multi-sequence casta new method for treating the children's proximal radioulnar joint fusion. *Food Sci Technol.* 2021;41(2): 603-610. doi: 10.1590/fst.33220.
- 16. El-Adl W. Two-stage double-level rotational osteotomy in the treatment of congenital radioulnar synostosis. *Acta Orthop Belg.* 2007;73(6):704-709.
- 17. Barrera-Ochoa S., Campillo-Recio D., Mir-Bullo X. Treating bilateral congenital radioulnar synostosis using the reverse Sauvé-Kapandji procedure. *J Hand Surg Eur Vol.* 2019;44(4):430-432. doi: 10.1177/1753193419827853.
- 18. Kawaguchi S., Kitamura M., Usui M. Proximal radioulnar synostosis treated with a free vascularised fasciofat graft report of two cases. *Hand Surg.* 2000;5(2): 161-164. doi: 10.1142/s0218810400000302.

- 19. Dawson H.G. A congenital deformity of the forearm and its operative treatment. *Br Med J.* 1912 Oct 52(2701): 833-835. doi: 10.1136/bmj.2.2701.833.
- 20. Hansen O.H., Andersen N.O. Congenital radio-ulnar synostosis. Report of 37 cases. *Acta Orthop Scand*. 1970;41(3):225-230. doi: 10.3109/17453677008991509.
- 21. Miura T., Nakamura R., Suzuki M., Kanie J. Congenital radio-ulnar synostosis. *J Hand Surg Br.* 1984;9(2):153-155.
- 22. Jia Y., Geng C., Song Z., Lv S., Dai B. Congenital unilateral proximal radioulnar synostosis: A surgical case report. *Medicine*. 2020;99(16):e19782. doi: 10.1097/MD.0000000000019782.
- 13. Prokopovich E.V., Konev M.A., Afonichev K.A., Prokopovich I.E., Kovzikov A.B., Nikitin M.S. et al. [Congenital radioulnar synostosis: symptom complex and surgical treatment]. *Travmatologiya, ortopediya i vosstanovitel'naya khirurgiya detskogo vozrasta* [Pediatric Traumatology, Orthopaedics and Reconstructive Surgery]. 2016;4(3):16-25. doi: 10.17816/PTORS4316-25. (In Russian).
- 24. Barik S., Farr S., Gallone G., Zarantonello P., Trisolino G., Di Gennaro G.L. Results after treatment of congenital radioulnar synostosis: a systematic review and pooled data analysis. *J Pediatr Orthop B*. 2021;30(6):593-600. doi: 10.1097/BPB.0000000000000841.
- 25. Rubin G., Rozen N., Bor N. Gradual correction of congenital radioulnar synostosis by an osteotomy and Ilizarov external fixation. *J Hand Surg.* 2013;38(3):447-452. doi: 10.1016/j.jhsa.2012.10.037.
- 26. Bolano L.E. Congenital proximal radioulnar synostosis: treatment with the Ilizarov method. *J Hand Surg Am*. 1994;19(6):977-978.doi:10.1016/0363-5023(94)90100-7.
- 27. Fujimoto M., Kato H., Minami A. Rotational osteotomy at the diaphysis of the radius in the treatment of congenital radioulnar synostosis. *J Pediatr Orthop.* 2005;25(5): 676-679. doi: 10.1097/01.bpo.0000161833.57409.bb.
- 28. Yammine K., Salon A., Pouliquen J.C. Congenital radioulnar synostosis. Study of a series of 37 children and adolescents. *Chir Main*. 1998;17(4):300-308.
- 29. Ramachandran M., Lau K., Jones D.H. Rotational osteotomies for congenital radioulnar synostosis. *J Bone Joint Surg Br.* 2005;87(10):1406-1410. doi: 10.1302/0301-620X.87B10.16445.
- 30. Hung N.N. Derotational osteotomy of the proximal radius and the distal ulna for congenital radioulnar synostosis. *J Child Orthop.* 2008;2(6):481-489. doi: 10.1007/s11832-008-0146-5.
- 31. Lin H.H., Strecker W.B., Manske P.R., Schoenecker P.L., Seyer D.M. A surgical technique of radioulnar osteoclasis to correct severe forearm rotation deformities. *J Pediatr Orthop.* 1995;15(1):53-58. doi: 10.1097/01241398-199501000-00012.
- 32. Murase T., Tada K., Yoshida T., Moritomo H. Derotational osteotomy at the shafts of the radius and ulna for congenital radioulnar synostosis. *J Hand Surg Am*. 2003;28(1):133-137. doi: 10.1053/jhsu.2003.50010.
- 33. Shingade V.U., Shingade R.V., Ughade S.N. Results of single-staged rotational osteotomy in a child with congenital proximal radioulnar synostosis: subjective and objective evaluation. *J Pediatr Orthop.* 2014;34(1): 63-69. doi: 10.1097/BPO.0b013e3182a00890.

- 34. Ahmad I., Khan S., Iqbal M.J., Khan M.A., Hakim A., Kamran K. Derotation osteotomy for congenital radioulnar synostosis. *KJMS*. 2015;8(1):117-121.
- 35. Kozhevnikov O.V., Kralina S.E. [Minimally invasive derotational osteotomy of the forearm bones in congenital radioulnar synostosis in children]. *Vestnik travmatologii i ortopedii im. N.N. Priorova* [N.N. Priorov Journal of Traumatology and Orthopedics]. 2017;24(3):43-45 (In Russian).
- 36. Castelló J.R., Garro L., Campo M. Congenital radioulnar synostosis. Surgical correction by derotational osteotomy. *Ann Chir Main Memb Super*. 1996;15(1):11-17. doi: 10.1016/s0753-9053(96)80019-6.
- 37. Mohammed T.C., Hicham A., Karima A., Lamyae C., Moulay-Abderahman A. Outcomes of derotation osteotomy at synostosis site for congenital radioulnar synostosis. *PAMJ Clinical Medicine*. 2019;1:31. doi: 10.11604/pamj-cm.2019.1.31.20680.
- 38. Ogino T., Hikino K. Congenital radio-ulnar synostosis: compensatory rotation around the wrist and rotation osteotomy. *J Hand Surg Br.* 1987;12(2):173-178. doi: 10.1016/0266-7681(87)90006-4.
- 39. Pasupathy B., Tholgappiyan T., Sureshbabu M. Congenital radio ulnar synostosis, analysis of functional outcome using double rotation osteotomy and osteotomy at synostosis site. *Int J Orthop Sci.* 2018;4(2):755-759. doi: 10.22271/ortho.2018.v4.i2k.110.
- 40. Pei X., Han J. Efficacy and feasibility of proximal radioulnar derotational osteotomy and internal fixation for the treatment of congenital radioulnar synostosis. *J Orthop Surg Res.* 2019;14(1):1-7. doi: 10.1186/s13018-019-1130-0.
- 41. Simcock X., Shah A.S., Waters P.M., Bae D.S. Safety and efficacy of derotational osteotomy for congenital radioulnar synostosis. *J Pediatr Orthop.* 2015;35(8): 838-843. doi: 10.1097/bpo.000000000000370.
- 42. Simmons B.P., Southmayd W.W., Riseborough E.J. Congenital radioulnar synostosis. *J Hand Surg.* 1983;8(6):829-838. doi: 10.1016/S0363-5023(83)80078-1.
- 43. Zhang Z.Q., Hu X.Y., Yuan H.J., Zheng P.F. Application of 3-Dimensional Printing Technology in Rotational Osteotomy of Congenital Radioulnar Synostosis. *J Hand Surg Am.* 2021;46(10):924.e1-924.e6. doi: 10.1016/j.jhsa.2020.12.004.
- 44. Cleary J.E., Omer G.E. Jr. Congenital proximal radio-ulnar synostosis. Natural history and functional assessment. *J Bone Joint Surg Am.* 1985;67:539-545.
- 45. Green W.T., Mital M.A. Congenital radio-ulnar synostosis: surgical treatment. *J Bone Joint Surg Am*. 1979;61(5):738-743.
- 46. Farzan M., Daneshjou Kh., Mortazavi S.M.J., Espander R. Congenital radioulnar synostosis: A report of 11 cases and review of literature. *Acta Medica Iranica*. 2002;40(2):126-131.
- 47. Shvedovchenko I.V. [Radioulnar synostosis]. In: [Orthopedics: National guideline]. Ed. by S.P. Mironov, G.P. Kotelnikov. Moscow; 2013. p. 182-183. (In Russian).

Authors' information

⊠ Yulia A. Fedorova

Address: 64-68, Parkovaya str., St. Petersburg, Pushkin, 196603, Russia

https://orcid.org/0000-0003-3842-2113

e-mail: julsigareva@gmail.com

Sergei V. Vissarionov — Dr. Sci. (Med.), Professor

https://orcid.org/0000-0003-4235-5048 e-mail: vissarionovs@gmail.com

Yaroslav N. Proschenko — Cand. Sci. (Med.) https://orcid.org/0000-0002-3328-2070

e-mail: yar-2011@list.ru

Sofia A. Gevorgiz

https://orcid.org/0000-0002-3489-3658 e-mail: sofiagevorgiz@gmail.com

Ekaterina A. Zakharyan — Cand. Sci. (Med.) https://orcid.org/0000-0001-6544-1657

e-mail: zax-2008@mail.ru