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Original article

The Role of Early Habilitation in Infants with Congenital Brachial Palsy

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SUMMARY

Congenital brachial palsy (CBP) can have long-term consequences such as deformities, contractures and growth imbalance of the affected arm and shoulder girdle that can lead to severe handicap.

The aim of the paper was to determine the effectiveness of early habilitation in infants with CBP.

This retrospective clinical study included 34 infants who were habilitated in the Physical Medicine and Rehabilitation Clinic, Clinical Center Niš, during the period from 2000 to 2017. The protocol consisted of passive and active-assisted range of motion exercises for the affected arm, neurodevelopmental treatment, warm packs and electrotherapy, starting from the 3rd week of life. At the Clinic, 20 sessions were conducted and parents were educated with handling and home exercise program. The infants were re-evaluated each month during the first year of life. The modification of the manual muscle test (MMT) was used to assess the muscle strength (grades 0-3). At 6 and 12 months of age, the outcome was defined as a full recovery (grade 3 of the affected muscles) and partial recovery (grades 1-2).

Male sex predominated (56%). The right arm was more commonly affected (65%). Birth weight was over 4000g in 65% of infants. Fifty-nine percent of infants had upper, 26.5% "extended" and 14.5% had complete root palsy. Associated injuries were noted in 10 infants. The habilitation started in the first month of life in 64.7% of infants. At 6 months of age, full recovery was achieved only in one infant, while at 12 months, 56% of infants were fully recovered (p < 0.05). The majority of fully recovered infants was from the group with the upper type of lesion (p < 0.05).

Early habilitation program is effective in infants with CBP. It is especially effective in the upper root palsy.

Key words: congenital brachial palsy, habilitation, physical therapy

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INTRODUCTION

Congenital brachial palsy (CBP) is the most common periferal neurological deficit presented at birth. In developed countries, the frequency ranges from 0.38 to 3 per 1,000 live-born children, while in underdeveloped countries it is estimated 5 per 1,000 live-born children (1-3). Congenital damage to the brachial plexus can have long-term consequences such as deformities and contractures (contracture of the glenohumeral joint, subluxation of the head of the humerus, radial dislocation, flexor contracture of the elbow, supinatory or pronatory contracture of the forearm, ulnar deviation of the wrist, finger contracture) leading to insufficient functions of the arm in daily activities and severe handicap (4, 5).

Clinically, congenital lesions of the plexus brachialis manifest at birth, and are classified into four types (4). The upper type or Erb's paralysis is the most common form. It occurs due to stretching, tearing or compression of the roots C5 and C6. The shoulder is adducted and internally rotated, the elbow extended and forearm pronated. The muscles affected by the lesion are: m. deltoideus, m. supraspinatus, m. teres minor, m. brachioradialis, m. biceps brachii and m. supinator. The "extended" Erb's palsy has a clinical picture as the upper type, and the lesion involves also the C7 root. Because of the radial nerve involvement, moderate elbow flexion is present, and m. triceps brachii, m. extensor carpi radialis, m. extensor carpi ulnaris and m. extensor digitorum communis are also affected. The clinical picture implies a "waiter's tip" position of the affected arm (the arm is adducted and in internal rotation in the shoulder joint, with extended elbow and pronated forearm, flexed wrist and extended fingers).

A complete lesion of plexus brachialis is the second most common. It is characterized by the complete paralysis of the arm due to the involvement of the branches of the entire plexus. The lower type or Klumpke's paralysis is very rare. It represents damage to the roots of C8 and T1 and only affects the hand. Ulnar and median nerves are affected. Damaged muscles are: mm. interossei, mm. eminentiae thenaris, mm. eminentiae hypothenaris, m. flexor carpi ulnaris and m. flexor digitorum profundus. If sympaticus branch originating from the root T1 is involved, Horner's syndrome (ptosis, myosis, enophtalmus, redness and anhydrosis) appears.

Based on the clinical picture and the characteristic position of the affected arm, a clinical examination allows making the diagnosis of CBP immediately after birth. Electromyoneurography (EMNG) can provide data on the degree of brachial plexus damage, and is useful in evaluation and prognosis of recovery (4). To exclude possible associated injuries (bone fractures, epiphyseolysis, subluxation and luxation of the joints), shoulder radiography is recommended. MRI is used to detect root avulsions, nerve compression or scarring (6).

Treatment starts in the first days after birth. Habilitation includes positioning and proper handling of the newborn, physical therapy (kinesiotherapy, thermotherapy, electrotherapy, parent education), orthoses and other aids (4). The first goal of the therapy is to prevent further damage, which is why early postnatal positioning is performed. The objectives of kinesiotherapy are to prevent the development of contractures and to improve the active muscular function by applying age-appropriate activities. The treatment process lasts continuously throughout childhood until the end of growth (1).

The prognosis depends on the severity of the lesion (degree of damage to the nerves) and on the applied and timely treatment. Complete recovery, recovery with minimal consequences in the form of muscular weakness and slight discoordination of the movement, as well as incomplete or no recovery with the advancement of contractures and small hand use, is possible. In cases where habilitation does not produce satisfactory results and the child does not achieve significant recovery by 4-6 months of age, microsurgery should be considered (2). Primary reconstructive surgical techniques include neurolysis, nerve grafting and nerve transfer (2). Secondary surgical interventions such as tenotomy, elongation, capsulotomy, transposition of the tendon may be indicated to help improve the function (1).

However, there is little evidence of the value of physiotherapy interventions in infants with CBP. Therefore, the aim of this study was to determine the importance of early habilitation in recovery of infants with congenital paralysis of plexus brachialis.

PATIENTS AND METHODS

The retrospective study included 34 infants who were habilitated at the Clinic of Physical Medicine and Rehabilitation, Clinical Center Niš, in the period from 2000 to 2017. Data on sex, maternal and obstetric history, birth weight, APGAR score, birth and associated injuries, birth asphyxia, type and side of the lesion, as well as data on initiation of therapy and conducted therapeutic procedures were taken from the medical histories of the infants.

The diagnosis of CBP of the newborn was set at the maternity ward after clinical examination based on the characteristic position of the affected arm, inability to perform active movements, full passive range of motion, reduced muscular tone, and asymmetric Moro reflex on the side of the lesion. If the infant experienced pain upon palpation of the clavicula and shoulder region, shoulder radiography was used to verify whether a clavicular or humeral fracture was sustained during delivery. Ultrasound examination of the neck and m. sternocleidomastoideus was performed in infants with head rotated opposite to the affected side and suspected torticollis. In neonates with perinatal asphyxia, ultrasound of the brain was indicated.

After the diagnosis of CBP was confirmed, the infants were referred to the Clinic of Physical Medicine and Rehabilitation for further habilitation treatment. Habilitation was carried out ambulatory for respondents from the Niš region, while respondents from distant regions, due to their distant living, were treated in stationary conditions.

At our Clinic, habilitation began in most infants in the 3rd week of life. Some infants from the distant regions started habilitation in their regional hospitals, but later they were referred to our Clinic due to postponed clinical improvement. In these infants, habilitation started at the latest in the 3rd month of life.

The therapeutic protocol began with the positioning of the paretic arm over the baby's chest. Kinesiotherapy consisted of exercises (passive and active-assisted) to increase the range of motion in the affected articular segments. A Bobath neurodevelopmental treatment was also conducted in order to stimulate motor development. Kinesiotherapy was conducted by educated physiotherapists once a day and parents were educated with handling and importance of performing exercises at home. As an introduction to kinesiotherapy, thermotherapy (paraffin pads) and electrotherapy (ascending galvanization and electrostimulation of paretic muscles) were applied. Paraffin pads were applied once a day for 20 minutes. During the ascendent galvanization, the anode was placed on the hand of the paretic arm, and the cathode on the shoulder region. The procedure lasted 20 minutes and was performed once a day. Electrical stimulations using exponential currents were applied to paretic muscles whose muscular strength was 0, 1 and 2 according to the modification of the Medical Research Council (MRC) scale. Electrical stimulation was performed until muscle fatigue once a day. From the 6th month of life, occupational therapy for the paretic arm was included in order to encourage the development of daily life activities. After 20 completed therapeutic procedures at the Clinic, infants were dismissed for home treatment

with advice that parents continue to perform their learned activities on a daily basis.

Prior to treatment, an evaluation of the muscular strength according to the modification of the MRC scale was performed. Because of the inability to perform active movements on demand in infants, grades from 0 to 3 were used. Grade 0 indicated the absence of contractions of the examined muscle, grade 1 the presence of contractions but without movement, grade 2 pointed to the limited range of movements in the joint, whereas grade 3 indicated the full range of movement. Our experienced physiotherapists tested the paretic muscles in supine, prone, sidelying and supported sitting with or without provocation. The following muscles were examined: m. deltoideus, m. latissimus dorsi, m. pectoralis major, m. infraspinatus, mm. rotatores interni, m. biceps brachii, m. triceps brachii, mm. pronatores, m. supinator, m. flexor carpi radialis et ulnaris, m. extensor carpi radialis et ulnaris, m. extensor digitorum communis and m. flexor digitorum superfitialis.

Control examinations were carried out at the Clinic every month in the first year of life of the respondents. In the sixth and twelfth months of the infant's life, a re-evaluation of the condition was made, whereby muscle strength of the paretic muscles was determined using a modified MRC scale. The obtained findings were compared with the initial, and the recovery was assessed as full (the affected muscles were rated grade 3), partial (affected muscles were with grade 2 or 1) or without improvement.

Electromyoneurography of the affected arm was performed during the habilitation process, only in infants in whom, despite treatment, recovery was postponed or partial.

The statistical program SPSS 21 was used for data processing. The obtained data on the monitored parameters were determined according to the methodology of descriptive and analytical statistics. Student's t-test and chi-square test were used. Significance levels in the results are indicated by: * p < 0.05.

RESULTS

The study group consisted of 34 infants, of which 56% were male. Twenty per cent of the children were from the Niš region, while the rest were coming from the south-eastern parts of Serbia. The right arm was more commonly affected (64.7%). Fifty-nine per cent of infants had upper, 26.5% "extended" and 14.5% complete root palsy.

Total number of newborns	34 (100%)	
Gender (male)	19 (56%)	
Affected arm (right)	22 (65%)	
Birth weight over 4000g	22 (65%)	
Mode of delivery		
• assisted (vacuum)	7 (20.5%)	
• breech	3 (8.8%)	
• spontaneous	24 (70.6%)	
Upper root palsy	20 (59%)	
Extended root palsy	9 (26.5%)	
Complete root palsy	5 (14.5%)	
Associated birth injuries	10 (29.4%)	
Clavicular fracture	6 (17.6%)	
Humeral fracture	1 (2.94%)	
Torticollis with sternocleidomastoid haematoma	2 (5.88%)	
Cephalhaematoma	1 (2.94%)	
APGAR score 1-7	22 (65%)	

Table 2. Muscle strength of the affected muscles grade 3, according to the modification of the manual muscle test, at baseline, at 6th and 12th month of life in infants with CBP

Examined muscle	baseline	6 months	12 months
Deltoid muscle anterior part	0%	42%	67.6%
Deltoid muscle lateral part	0%	26.4%	50%%
Deltoid muscle posterior part	0%	8.8%	38.2%
Infraspinatus muscle	0%	11.7%	35.3%
Latissimus dorsi muscle	0%	17.6%	47%
Pectoralis major muscle	2.94%	47%	88.2%
Internal rotators	2.94%	29.4%	53%
Biceps brachii muscle	2.94%	52.9%	73.5%
Triceps brachii muscle	23.5%	85.3%	91.1%
Supinator muscle	0%	8.8%	26.5%
Pronator muscles	26.4%	70.5%	94.1%
Flexor carpi radialis muscle	58.8%	91.1%	91.1%
Flexor carpi ulnaris muscle	61.8%	91.1%	91.1%
Extensor carpi radialis muscle	38.2%	79.5%	85.3%
Extensor carpi ulnaris muscle	38.2%	79.5%	85.3%
Extensor digitorumcommunis muscle	79.5%	91.1%	94.1%
Flexor digitorumsuperfitialis muscle	67.6%	91.1%	91.1%

		Complete recovery		Partial recovery	
Gender	male	11	32.3%	8	23.5%
	female	8	23.5%	7	20.5%
Body mass	>4,000g	12	35.3%	10	29.4%
	< 4,000g	7	20.5%	5	14.7%
Birth risk factors	present	6	17.6%	4	11.7%
	absent	11	32.3%	13	38.2%
APGAR	0-3	2	5.88%	1	2.94%
	4-7	13	38.2%	9	26.4%
	8-10	5	14.7%	5	14.7%
Affected arm -	right	5	14.7%	1	2.94%
	left	14	41.2%	14	41.2%
Type of lesion	upper*	15	44.1%	5	14.7%
	extended	3	8.82%	6	17.6%
	complete	1	2.94%	4	11.7%
Asphyxia	present	13	38.2%	11	32.3%
	absent	7	20.5%	5	14.7%
Beginning of - habilitation -	1. month	12	35.3%	9	26.4%
	2. month	5	14.7%	4	11.7%
	3. month	2	5.88%	2	5.88%
Duration of habilitation*	6 months	1	2.94%	33	97%
	12 months	19	55.8%	15	44.1%

Table 3. The influence of risk factors, the beginning and duration of habilitation on the outcome at 12th month in infants with CBP

*p < 0.05

All newborns were termed. The birth weight over 4,000 grams had 22 newborns, of which 5 were weighting over 4,500 grams. Birth asphyxia was present in 22 newborns. The mode of delivery was spontaneous in 70.6%, assisted in 20.5% and breech in 8.8%. Associated birth injuries had 10 newborns: 6 fracture of the clavicle, 1 fracture of the humerus, 2 myogenic torticollis with haematoma in m. sternocleidomastoideus and 1 cephalhaematoma (Table 1).

The habilitation started in the first month of life in 64.7% of infants. In the sixth and twelfth months of the life of the subjects, improvements in muscle strength of affected muscles were noted. After six months of habilitation, the greatest progress was achieved in the following muscles: m. deltoideus pars clavicularis (anterior

part), m. pectoralis major, m. biceps brachii, m. triceps brachii, mm. pronatores and m. extensor carpi radialis et ulnaris (the percentage of infants with grade 3 increased by 40%). The muscles with the weakest recovery, even after 12 months were: m. deltoideus pars spinata (posterior part), m. supinator, m. infraspinatus, m. latissimus dorsi (less than 50% of infants had grade 3 after 12 months of habilitation) (Table 2).

Table 3 shows the influence of risk factors, the beginning and duration of habilitation on the outcome at 12th month. The duration of habilitation significantly affected the treatment outcome. At 6 months of age, full recovery was achieved only in one infant, while at 12 months, 56% of infants were completely recovered (p < 0.05). Full recovery was achieved in 15 infants with

upper lesion type, 3 with "extended" and 1 with complete lesion type. The majority of fully recovered infants (79%) was from the group with the upper type of lesion (p < 0.05).

Differences in the onset of habilitation (first, second or third month of life) did not show a statistically significant effect on the final outcome of habilitation, although the highest number of fully recovered children were from the group that started habilitation in the first month. Gender, APGAR score, the presence of birth risk factors and associated congenital injuries did not prove to be a statistically significant predictor of full recovery. Body weight above 4,000 grams did not have an effect on the outcome of treatment, nor did perinatal asphyxia (Table 3).

DISCUSSION

In this retrospective study, the significance of early habilitation in infants with CBP was investigated. Our results showed that infants who started the habilitation in the first month improved progressively, with improvement already in the 6th month of life. By the end of the first year, slightly more than half of infants reached full recovery (56%).

The complete recovery rates in children with CBP vary between the studies from 13% to 80% (7). The recovery depends on the degree and type of the brachial plexus injury, applied treatment and follow-up period. The four types of nerve injury are described such as avulsion, rupture, neuroma and neuropraxia (8). Nerve avulsions are preganglionic and the nerve is torn away at its point of attachment to the spinal cord. Nerve rupture indicates tearing (partial or complete) of the nerve distal from its site of attachment. A traumatic neuroma can be considered as scar tissue that may lead to nerve compression. Neuropraxia due to solely nerve stretching can be described as a transient loss of motor or sensory function resulting from the blockage of nerve conduction, usually lasting an average of 6 to 8 weeks before full recovery.

Newborns with neuropraxia can achieve 90% to 100% recovery. Gilbert et al. reported on a recovery rate of 80% to 96% in infants in whom clinical improvement started in the first two weeks (9). In infants with axonot-mesis, the largest number, 55%, is recovered in the period from 6 to 12 months, at the latest until the 18th month of the child's life (1). In contrast, infants with nerve root avulsion or proximal nerve tears have no potential for recovery (10). In these children, who do not show

any recovery by 3 to 5 months, surgical exploration of the brachial plexus should be considered (11).

In newborns with CBP, muscle strength can be assessed using the Active Movement Scale, the Toronto Test, the Mallet Modified Scale and the Medical Research Council Scale (3, 12, 13). We used the modification of the Medical Research Council Scale according to Gilbert and Tassin, which can be applied in the youngest age (4). Instead of grades 0-5, it uses grades 0-3 because infants cannot perform movement on demand or against resistance.

Nerve conduction studies can identify nerve avulsion injuries, nerve root rupture and neuropraxia (8). The use of EMG in routine evaluation after CBP remains controversial. There is a high probability of false negative findings in the neonates and false positive findings in a few months post-term (3). There is also a discrepancy between clinical and EMG findings. Van Dijk et al. reported that small axonal size in neonates could reduce the time to complete the process of denervation and the start of re-innervation in comparison to adults (14). In our study, EMNG was performed only in infants in whom, despite treatment, recovery was postponed or partial. Infants in whom the severe damage of the brachial plexus was verified were referred to a pediatric neurologist or neurosurgeon for further evaluation.

Our study has shown that the majority of fully recovered infants (79%) had upper root palsy. Habilitation was less successful in infants with other types of lesions. Children with complete brachial plexus palsy showed only partial improvement at 12th month. Our results can be compared with other studies. In the study of Joksimović et al., infants with the upper type of brachial palsy were completely recovered, while in the infant with the lower type, only marked improvement was registered (15). Infants with the complete lesion have 2 times greater possibility of developing paralysis compared to the upper type of lesion (16).

The application of early diagnostics and physical therapy is an important factor in preventing permanent consequences and handicap due to the reduced function of the affected arm in infants with CBP (17). In our study, although all infants progressed, and habilitation began at the latest in the 3rd month of life, the largest number of fully recovered infants was from the group that started treatment in the first month of life (15 out of 19 infants).

The time when the recovery of function occurs is an important prognostic factor. Patients exhibiting activity in deltoideus, biceps brachii and triceps brachii muscle until the 4th month of life have the greatest chance of achieving complete recovery (18). In the 6th month of life, more than half of our respondents achieved significant functional activity in biceps brachii (53%) and triceps brachii muscle (85%). For the deltoideus muscle, the highest percentage of infants reached the improvement of the clavicular part (42%), somewhat smaller (26%) of the acromial, and the smallest (8%) of the spinata part. Given that 56% of infants have achieved complete recovery, it can be assumed that achieving functional activity in these muscles early after birth increases the chance for complete recovery.

The degree of brachial plexus injury at birth is the reason for the different success of therapy, according to the study of Papazian et al. (19). There is a high variability in treatment success, ranging from 4% to 93% (4). This variability in treatment effectiveness results from the lack of complete documentation in each research, as well as insufficient long-term follow-up period of children and different methods used in evaluation (4). Studies that do not follow children until the 3rd year of life can underestimate the remaining deficits of the affected arm in everyday activities (20).

Predisposing factors for the development of congenital palsy of plexus brachialis can be divided into those related to the fetus (neonatal), to factors related to the mother and factors related to the delivery (4).

The most important neonatal factor is the birth weight over 4,000 grams (1). The American College of Obstetricians and Gynecologists pointed out that neonates over 4,500 grams have 45 greater chance of developing CBP (4). In our study, 22 newborns had a body mass over 4,000 grams. This data confirms that macrosomy of the fetus has significance in the onset of lesion of the plexus brachialis. Our study has shown that it had no effect on the final outcome of the treatment. Infants with a weight of 4,000 grams did not show less probability of achieving complete recovery.

Maternal risk factors include diabetes mellitus (which leads to macrosomia), obesity (which leads to weak muscle of the anterior abdominal wall and prolonged delivery), age over 35 years, and anatomic pelvic characteristics (fetal-maternal disproportion) (1).

Perinatal risk factors relate to disorders of the position and accommodation of the fetus, and the use of the obstetric interventions (vacuum extraction, forceps) that can lead to shoulder dystocia behind the symphysis and clavicular or humeral fractures (1). In our respondents, perinatal risk factors were present in only 10 children. These data indicate that the presence of perinatal risk factors does not necessarily lead to the lesion of the plexus brachialis, but also that their absence is not a guarantee that the lesion will not occur.

In our study, only a third of infants had associated injuries suggesting that newborns experienced some kind of birth trauma (clavicular fracture was present in 6 cases, a fracture of the humerus in 1, a sternocleidomastoid muscle haematoma and torticollis in 2, and a cephalhaematoma in 1 case). Our results point out to the possibility of the effects of prenatal factors on the occurrence of damage to brachialis plexus (umbilical cord wrapped around the shoulder, neck and arm, position of the fetus) (4, 7).

The majority of our respondents, as much as 65%, had asphyxia at birth. There is a frequent occurrence of asphyxia in newborns with CBP. Blacke et al. in their study showed asphyxia as one of the risk factors, both for the appearance of the lesion of the plexus brachialis, as well as for its permanent damage (21). In our group of subjects, although it was present in 22 infants, asphyxia at birth did not have an impact on the course and the final outcome of the recovery.

CONCLUSION

With the application of habilitation, as early as the first month of the infant's life, a partial recovery was achieved already in the sixth month of life, and by the end of the first year, full recovery was achieved in 56% cases. Depending on the characteristics of the infant (body weight) or complications that occurred during delivery (obstetric interventions, birth injuries, asphyxia), habilitation measures are of the same importance in achieving recovery. Habilitation is particularly effective in the upper type of lesion, where up to 79% is recovered by the end of the first year. It is less successful in the infants with "extended" type, where complete recovery is achieved in a half of infants. Habilitation is the least effective in infants with a complete type of lesion, of which from five infants only one recovered completely to the end of the first year.

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Uloga rane habilitacije u lečenju odojčadi sa kongenitalnom paralizom pleksusa brahialisa

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SAŽETAK

Kongenitalna paraliza brahijalnog pleksusa (KBP) može imati dugotrajne posledice kao što su deformacije, kontrakture i zaostajanje rasta ruke i ramenog pojasa, što može dovesti do ozbiljnog hendikepa deteta.

Cilj rada bio je utvrđivanje značaja rane habilitacije u oporavku odojčadi sa KBP.

Retrospektivnom studijom obuhvaćena su 34 odojčeta koja su habilitovana na Klinici za fizikalnu medicinu i rehabilitaciju Kliničkog centra u Nišu, u periodu od 2000. do 2017. godine. Terapijski protokol je podrazumevao primenu pasivnih i aktivno-potpomognutih vežbi za paretičnu ruku, neurorazvojni tretman, termoterapiju i elektroterapiju, počevši od treće nedelje života odojčeta. Na Klinici je sprovedeno 20 terapijskih procedura, pri čemu su roditelji edukovani za vežbanje kod kuće. Reevaluacija je obavljena svakog meseca u toku prve godine života. Za procenu grube mišićne snage koriščena je modifikovana skala manuelnog mišićnog testa (ocene od 0 do 3). U 6. i 12. mesecu života odojčeta, oporavak je definisan kao potpun (ocena 3 za paretične mišiće) i parcijalan (ocena 1-2).

Odojčad muškog pola bila je zastupljenija (56%), kao i zahvaćenost desne ruke (65%). Porođajnu težinu veću od 4000 grama imalo je 65% odojčeta. Gornji tip paralize bio je zastupljen kod 59% odojčadi, "proširen" u 26,5%, a u 14,5% kompletan tip. Udružene porođajne povrede je imalo 10 novorođenčeta. Habilitacija je započela u prvom mesecu života kod 64,7% odojčadi. Nakon 6 meseci potpuni oporavak je postignut samo kod jednog odojčeta, dok je nakon 12 meseci, 56% odojčeta bilo potpuno oporavljeno (p < 0,05). Većina potpuno oporavljenih bila je iz grupe sa gornjim tipom lezije (p < 0,05).

Rana habilitacija je efikasna u lečenju dece sa KBP. Najbolji oporavak se postiže kod gornjeg tipa lezije pleksusa brahijalisa.

Ključne reči: kongenitalna paraliza pleksus brahijalisa, habilitacija, fizikalna terapija