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Research Article

The Role of Early Family-Centered Therapy Program in Infants With Brachial Plexus Birth Palsy

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Doğumsal Brakiyal Pleksus Felçli Bebeklerde Aile Odaklı Erken Terapi Programının Rolü

ABSTRACT

Objective: *The purpose of this study is to evaluate the effectiveness of a family-centered early exercise program in infants with Narakas Stage 1 brachial plexus birth palsy.*
Method: *The data of the infants with brachial plexus birth palsy followed up at the physical medicine and rehabilitation outpatient clinic of a tertiary pediatric research center were retrospectively investigated. Infants with Narakas Stage 1 brachial plexus birth palsy according to the Narakas classification were included in the study. Sixty infants were assessed using a passive-active range of motion (ROM) and active movement scale at first clinic visit and every month until they became 12 months old.*
Results: *Cases were divided into two groups (Group I, n:32) or (Group II, n:28) according to their referral times to the outpatient clinics of physical medicine and rehabilitation. In both groups, a significant improvement was observed in the ROM and muscle strength of shoulder, elbow flexion, and forearm supination at the 3rd, 6th and 12th-month assessments. At 12th months intergroup differences were detected in shoulder abduction muscle force assessments ($p<0.05$) and active ROM measures of shoulder abduction-internal rotation-external rotation and forearm supination ($p<0.05$).*
Conclusion: *Family-centered early therapy program is effective against complications that may occur in the first year of infants with Narakas Stage1 brachial plexus birth palsy. In infants with brachial plexus birth palsy, not only neurological improvement should be focused, but also regular follow-ups should be made regarding complications.*

Keywords: *Brachial plexus birth palsy, brachial plexus, physiotherapy*

ÖZ

Amaç: *Bu çalışmanın amacı, aile odaklı erken terapi programının Narakas evre 1 olan doğumsal brakiyal pleksus felçli infantlar üzerindeki etkinliğini değerlendirmektir.*
Yöntem: *Üçüncü basamak pediyatrik araştırma merkezinin fiziksel tıp ve rehabilitasyon polikliniğinde takip edilen doğumsal brakiyal pleksus felçli infantların verileri retrospektif olarak incelendi. Çalışmaya Narakas sınıflamasına göre Narakas Evre 1 brakiyal pleksus doğum felçli bebekler dahil edildi. Altmış bebek, pasif-aktif hareket açıklığı (EHA) ve aktif hareket ölçeği ilk klinik ziyareti ve sonrasında 12 aylık olana kadar her ay değerlendirildi.*
Bulgular: *Olgular, fiziksel tıp ve rehabilitasyon kliniğinde polikliniğine sevk zamanına göre iki gruba (Grup I, 32 olgu) (Grup II, 28 olgu) ayrıldı. Her iki grupta da 3., 6. ve 12. ay değerlendirmelerinde omuz, dirsek fleksiyonu ve önkol supinasyonunun EHA ve kas gücünde anlamlı iyileşme gözlendi. Omuz abdüksiyon kas kuvveti değerlendirmelerinde ($p>0,05$) ve omuz abdüksiyon-iç rotasyon-dış rotasyon ve önkol supinasyon aktif EHA ölçümlerinde 12. ay grupları arasında farklılıklar vardı ($p<0,05$).*
Sonuç: *Aile odaklı erken terapi programı Narakas evre 1 doğumsal brakiyal pleksus felçli infantlarda hayatın ilk 1 yılında oluşabilecek komplikasyonlara karşı etkilidir. Doğumsal brakiyal pleksus felçli bebeklerde sadece nörolojik iyileşmeye odaklanılmamalı, aynı zamanda komplikasyonlar ile ilgili düzenli takipler yapılmalıdır.*

Anahtar kelimeler: *Doğumsal brakial pleksus felci, brakial pleksus, fizyoterapi*

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INTRODUCTION

Brachial plexus birth palsy (BPBP) is an upper extremity paralysis that occurs due to traction injury of the brachial plexus during birth [1]. Various data were obtained at different times and locations in terms of its incidence. Its incidence has been reported at a rate ranging from 0.42-4 per 1000 live births [2,3]. As per Narakas' classification, the most obstetrical brachial plexus injuries are classified as type 1: Erb's palsy involving the spinal levels of C5 and C6. Studies have shown that the course of narakas type 1 BPBP leads a better course [4,5]. Howover some of these infants with a seemingly complete neurologic recovery will develop a shoulder contracture or subluxation during growth [6]. Several studies submitted their functional results however scheduling-planning of exercise programs and their contributions have not been presented in detail [4, 7, 8]. For this reason, this study was planned to investigate the effectiveness of the early exercise program applied on infants with narakas grade 1 brachial plexus birth palsy.

MATERIAL and METHODS

Newborns with diagnosis of the BPBP newborn infants referred to the physical medicine and rehabilitation (PMR) outpatient clinic of Izmir Dr. Behçet Uz Pediatric Diseases and Surgery Training and Research Hospital over a three-year period were included in this study. The infants were identified by retrospectively investigating a prospective BPBP record at a single tertiary pediatric research hospital.

Patients were classified using Narakas classification [9]. All nonsurgical candidates (Narakas class 1) were included in the study. The patients who had another comorbid neurological problem, fracture on the affected arm, lost to follow-up (<12 mo) and surgical candidates were excluded from the study (*n*=4). Sixty-four infants constituted the study sample. At each clinical visit, patients underwent standardized physical examination every month with use of the Active Movement Scale (AMS) and measurements of passive and active range of motion of the elbow and shoulder. Recovery of antigravity elbow flexion was defined as AMS ≥5 for elbow flexion. Our work was followed up regularly every month until one year but was given statistical data for the first, 3. months, 6. months and 12. months of age.

Since 4 patients had biceps muscle force <3 AMS when they were 4-5 months old, these children were referred to surgery and excluded from the study. A retrospective search of this prospective database identified 60 patients.

Demographic information including age at the time of referral for PMR outpatient clinic for initial functional evaluation, gender and affected side was recorded. Subjects were divided into two groups (group I, n:32) or (group II, n:28) according to PMR referral time. The group I received a family-centered therapy program starting from the 3 weeks of life (Early referred to PMR), whilst the group II received this program the after 3 weeks of life (Delayed referred to PMR). Thirty two (%53) patients were referred within the 3 weeks after delivery, while 28 (%47) applied after the first 3 weeks of life.

The families were trained by a physiotherapist at the hospital through a visual and applied training program, and then the parents were tested. The patients are evaluated by the same physiatrist for sustained. Within the scope of this program, a few days after birth, the patients were recommended to support the arm in the neutral position for 10 days, and after 10 days the joint movements of the paralyzed limb were initiated. Parents were taught a home-based range of motion exercise program, and guided by monthly therapy sessions. Very gentle passive range of motion (PROM) were given every waking hour for 10 repetitions for shoulder flexion, abduction-external rotation, elbow flexion and extension, forearm supination and pronation and wrist flexion and extension. One month aftery delivery passive joint movements were first shown to the parents and progressed towards active reinforcement, active positioning and strengthening exercises against gravity in the periodic evaluator lights. Passive joint movements were first shown to the parents, and proceeded towards active assistive

and strengthening exercises against gravity in the periodic evaluative directions. In order to increase the awareness of the child's paretic extremity, the family were taught initiatives that could induce tactile stimulation, such as contacting materials with different tissue surfaces along the child's affected arm and transferring weight to the affected arm. In order to increase the awareness of the child's paretic extremity, the family were taught initiatives that could induce tactile stimulation, such as contacting materials with different tissue surfaces along the child's affected arm and weight-bearing exercises. After the 4 th; the exercises in the previous stages were continued. Bimanual activities were encouraged to avoid neglect of the limb involved [10].

Assessment criteria: Narakas classification; this classification consists of four groups: Group I C5-6 shoulder and biceps paralysis, Group II C5-7 shoulder, biceps and forearm extremities, Group III C5-Th1 complete paralysis of the extremity and Group IV C5-Th1 complete paralysis of the extremity accompanying Horner syndrome [9]. The Active Movement Scale (AMS): AMS is a method recommended for the evaluation of patients with brachial palsy from the newborn period to the age of one [11,12]. At each visit functional recovery is assessed with a special grading system based on" the AMS" as below. The Active Movement Scale (AMS) is used to objectively examine the activation of UE muscle groups. Scores for UE joint motions are graded on a 0-7 point scale based on the percent of active motion observed within the available PROM. Active motions performed with minimize impact of gravity scored from 0-4 and motions performed against gravity are scored from 5-7 [11,12] (Table 1). Range of motions (ROM) involving flexion-extension and abduction adduction and internal and external rotation of the shoulder, flexion, extension and supination of the elbow are measured by goniometer and then recorded.

Ethical Approval: All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee as well as the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The study was approved by the medical local research ethics committee of Izmir Dr. Behçet Uz Pediatric Diseases and Surgery Training and Research Hospital (26.12.2013/No:1339918/80).

Statistical Analysis: Statistical evaluation was performed using PASW Statistics for Windows, Version 18.0. (SPSS Inc. Chicago, USA, Released 2009). The Student t-test was used for independent groups to analyze data that complied with a normal distribution. A paired sample t-test was used for dependent group analysis $p<0.05$ was considered to be statistically significant.

RESULTS

According to Narakas classification, 64 patients asessed as grade I cases were included in this study. Since biceps muscle strenght of 4 patient was AMS <3 when they were 4-5 months old, these children were referred to surgery and excluded from the study. So a total of 60 patients were included in the study. There were a total of 32 patients in Group I (15 girls, 17 boys; mean age 7.4±day, mean birth weight 3420±287 grams) and 28 patients in group II (13 girls,15 boys; mean age 37.8±0.5 day, mean birth weight 3485±216 grams). The general characteristics of both groups are presented in Table 1. At baseline, there were no significant differences between two groups.

İntragroup statistical analysis inside the groups: In terms of active ROM, while a statistically significant improvement was achieved in both groups between

Table 1. Hospital for sick children muscle grading system.

Observation Muscle grade	
Gravity eliminated	0
No contraction	1
Contraction, no motion	2
Motion<1⁄2 range	3
Motion> 1⁄2 range	4
Full motion	
Against gravity	5
Motion<1⁄2 range	6
Motion> 1⁄2 range	7
Full motion	

Table 2. Characteristics of patients of BPBP.

n=60	Group I (n=32)	Group II (n=28)	p
Age (days)			
Mean±SD	7.4±2.0	37.8±5.0	<0.001
Sex, n (%) *			
Girl	15 (46.9)	13 (46.4)	
Boy	17 (53.1)	15 (53.1)	0.603[2]
Affected side, n (%) *			
Right	17 (53.1)	13 (46.9)	
Left	15 (46.9)	15 (53.1)	0.603[2]
Birt weight (g)			
Mean±SD	3420±287	3485±216	0.515[1]

**Row percentage, [1]t-test, [2]chi square-test*

1st-3rd months, 3rd-6th months and 6th-12nd months in shoulder abduction-flexion, external-internal rotation and elbow flexion and forearm supination (p<0,001). Active ROM measures of the patients are shown in Table 3. A significant muscle strength improvement was observed in both groups between 1st, 3rd, 6th, and 12nd months in shoulder abduction, shoulder flexion, elbow flexion and forearm supination (p<0.001). The active movement scale (AMS) measures of the patients are shown in Table 4.

Statistical analysis between the groups: statistically significant difference was observed between the

Table 3. Hospital for sick children muscle grading system in patients with BPBP.

	Birth		3 months		6 months		12 months	
n=60	Group I (n=30)	Group II (n=30)	Group I (n=30)	Group II (n=30)	Group I (n=30)	Group II (n=30)	Group I (n=30)	Group II (n=30)
Shoulder								
Abduction[1]	2.5±0.5	2.7±0.4	3.8±0.4	3.8±0.5	5.3±0.5	5.0±0.6	6.5±0.5[2]	6.2±0.4[2]
Flexion[1]	2.4±0.6	2.6±0.3	4.6±0.7	4.5±0.7	5.6±0.4	5.5± .5	6.6±0.4	6.4±0.5
Elbow								
Flex[1]	0.8±0.6	1.1±0.7	3.5±0.5	3.4±0.5	5.5±0.5	5.5±0.5	6.6±0.4	6.5±0.5
Forearm								
Supination[1]	0.5±0.5	0.7±0.6	2.8±0.4	2.9±0.5	5.5±0.7	5.4±0.5	6.5±0.5	6.4±0.5

[1]: Statistically significant differences according to the birth value inner group I and II evaluations (p<0.001).
[2]: p<0.05 for each between group evaluations.

Table 4. Measurements of active range of motion in patients with BPBP.

	Birth		3 months		6 months		12 months	
n=60	Group I (n=32)	Group II (n=28)	Group I (n=32)	Group II (n=28)	Group I (n=32)	Group II (n=28)	Group I (n=32)	Group II (n=28)
Shoulder								
Abduction[1]	13.7±3.3	15.7±5.4	37.0±4.4	36.6±4.7	62.2± .0	59.2±7.4	103.7±27.6[2]	88.0±21.6[2]
Ext rot[1]	0	0.3±2.6	18.1±1.9	17.7± .3	24.9±1.9	23.9± .1	33.4±9.1	30.5± 6.5[2]
Int rot[1]	36.0±4.2	39.2± .2	59.1±3.4	57.5± .0	76.3±3.5[2]	73.5±4.5[2]	79.0±2.4[2]	76.0±4.5[2]
Flexion[1]	20.3±3.6	23.7±3.0	48.8±4.3	46.6±9.4	67.3±12.7	73.3±22.1	92.8±21.4	87.1±2.5
Elbow								
Flexion[1]	0.9±1.5	1.9±3.0	65.7±4.1	63.2±5.4	78.2±3.8	76.4±3.6	103.7±5.6	101.0±6.8
Forearm								
Supination[1]	1.1±2.0	1.6±2.3	31.6± .6	29.2±5.3	68.1±7.2	65.4±8.6	79.8±6.6[2]	74.2±7.4[2]

[1]: Statistically significant differences according to the birth value inner group I and II evaluations (p<0.001).
[2]: p<0.05 for each between group evaluations.

groups At 12nd months in shoulder abduction muscle force assessments and (p>0.05) shoulder abduction-internal rotation-external rotation and 6th months in shoulder internal rotation (p<0,05).

All infants regained antigravity elbow flexion (AMS≥5) at a median of 6 months. Elbow outcomes remained unaffected by delayed referral to PMR. Early referral PMR clinic were associated with an improved shoulder outcome. As for range of passive motion two children had reduced shoulder external

rotation and observed revealed posterior glenohumeral dislocation, two supination contracture and the trumpet sign was present in 2 children at 12 months of age in group II.

DISCUSSION

The Group I received a family-centered early therapy program starting from the 3 weeks of life, whilst the Group II took this program after 3 weeks of life. The parents of 28 children were unable to early bring their children to the hospital, because they were living in the rural or remote areas. This study proved that regarding Narakas 1 BPBP patients, any significant difference did not exist between the groups who had, and had not started exercise programme at an early stage as for functional improvement in elbow. In all the patients who early initiate exercise program nearly normal shoulder functions were found 12 months old. Shoulder complications developed in the patients during their follow-ups who did not the initiate exercise program early. Early referral for physical therapy has been observed to be beneficial to children with BBPI.

Meta-analysis-based results revealed that the prognosis of Narakas stage 1 cases have a more improved prognosis [13]. However, it should be kept in mind that approximately 20-30% of patients with C5-C6 root involvement may develop permanent deficiencies on the upper limb [1]. A physical therapy program is the recommended treatment for patients with BPBP at the first stage. The main goal of physical therapy program is to prevent the development of contractures in the period when improvement of passive joint movement, the flexibility of joints and muscle strength are expected and healing of the plexus. In many studies; the effectiveness of primary conservative treatment in infants with BPB has been investigated, but a complete conclusion could not be achieved [8,14]. Some authors suggested that exercises did not have much effect on recovery [5]. However, there are limited studies about the time-planning of exercise programs for the patients with brachial plexus birth palsies.

It is generally believed that most BPBP patients will recover spontaneously [15]. However, many studies have overestimated the spontaneous functional recovery rate, as reported in the systematic review of Pondaag et al [16]. Hamzat et al. [17], reported that some infants with BPBP were referred for physiotherapy more than 6 months after birth. In this study, it was observed that some narakas 1 cases were neglected and were not directed in the early period. Parents who may not have appreciated the significance of the disorder can delay their appointments for various family-social-economic reasons and difficulties in accessing PMR clinic. In some cases, the movements of the biceps and deltoid muscles that could never reach a sufficient level; cause complaints induce optimistic expectations in the parents of the patient and the infant is not brought for control.

DiTaranto et al [18] evaluated 91 infants with BPBP treated conservatively for 2 years. In 69% of the patients with a typical Erb's palsy functions of biceps, deltoid, triceps, and wrist recovered well by 6 months, and by 2 years, only mild shoulder range of motion was only mildly restricted. Therapy consisted of active, active-assisted and passive range of motion exercises. Azzi et al reported an association between delayed referral Narakas class 1 BPBP the the obstetric brachial plexus clinic and shoulder outcome. They stated that the early referral to the PMR clinic crucially prevents joint contractures. However, the frequency and intensity of physical therapy were not taken into account in this study [19].

Ultrasound is recommended to detect infantile shoulder dislocation [20]. Therefore, in our study, we evaluated suspicious patients with ultrasound. Dahlin et al; described six of 82 patients with brachial plexus birth palsy who developed a posterior dislocation of the shoulder during the first year of life [21]. In our study, 5 patients were referred for baseline shoulder ultrasonography at 12 months of age and posterior glenohumeral dislocation was revealed in 2 patients aged> 3 weeks who applied to PMR clinic.

A patient who detected internal rotation contracture of the shoulder in the 9th months of age has used a SUP-ER orthosis for three months. An

increase in the ROM of the passive shoulder external rotation was observed at the 12th month. Weekly kinesiology taping added to stretching exercises was applied to 2 patients who developed supination strain in 9 months. The kinesiotype of the patients was renewed every week until 12 months. The supination strain of these patients was resolved at 12 months. However, supination contracture and trumpet sign were observed in 2 different patients at 12 months. No relationship was found between rotation movements of the forearm and the onset time of biceps recovery. The age at which biceps function recovers is not a reliable predictor of forearm rotation.

Exercises increase the sensory input which results in increasing plasticity. The development of plasticity can be precipitated with excercises initiated at an early stage and repeated frequently, and regularly [22,23]. In the first months of neonatal life, passive joint movement exercises and stretches performed depending on the number and set of motion repetitions performed with the affected upper extremity may play a role in preparing the environment for regeneration by preventing secondary changes that may ocur during joint movements. In our study, it was shown that secondary deformities were prevented by shoulder positioning and range of motion exercises taught to the family from the birth. It is important to perform regular measurements in children with birth-related brachial plexus palsy in order to follow the clinical course adequately and detect and take precautions against future deformities.

Our results indicate significant improvement in ROMs, and the function to the upper extremity functions by means of an early exercise program. The newborn with BPP should be referred to PMR at an early stage and then followed-up regularly. Focus should not be placed only on neurological improvement but the patients should also be monitored regarding complications. Since the rate of spontaneous recovery tends to be less than the previously reported literature data, early diagnosis and regular follow-up should be performed by a qualified healthcare provider who is aware of the complications and long-term effects of this condition.

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