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ORIGINAL RESEARCH

Effect of reaching training combined with electrical stimulation in infants with brachial plexus palsy: a single subject design

Efeito do treino de alcance combinado com estimulação elétrica em lactentes com paralisia braquial perinatal: estudo experimental de caso único

Efecto del entrenamiento de alcance combinado con electroestimulación en los lactantes con parálisis braquial perinatal: un estudio experimental de caso único

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ABSTRACT | This study aimed to evaluate the effect of reaching training combined with functional electrical stimulation (FES) on active upper limb movement in infants with perinatal brachial palsy (PBP). Experimental single-case A-B design with follow-up. Two infants participated in the study, one girl of 7 months and one boy of 10 months of age. Data on infant's upper limb function (Active Movement Scale) were documented weekly. After six baseline assessments (A), 3 times per 2 weeks, intervention consisted of applying FES Neurodyn® to the deltoid muscle combined with objects reach training, encouraging abduction and anterior shoulder flexion above 90º, for six weeks, totaling 15 visits (B). The infants were reevaluated twice after a period of 15 and 30 days without intervention (follow up). The total score obtained on each evaluation day was plotted graphically. Electrostimulation was well accepted, with no side effects. Both infants showed improvement in the active movement of the affected upper limb after the intervention and the gains were maintained at follow-up, an increase of 9 and 7 points for infants 1 and 2, respectively. Six weeks of intervention resulted in individual changes in infant's upper limb function, mainly increased active range of motion in shoulder flexion and abduction and elbow flexion. The results of this study suggest the use of electrical stimulation as an adjunct to training the use of the affected upper limb of infants with PBP.

Keywords | Brachial Plexus Neuropathies; Electrical Stimulation; Physical Therapy Specialty.

RESUMO | O objetivo deste estudo foi avaliar o efeito do treino de alcance combinado à estimulação elétrica funcional (FES) na movimentação ativa do membro superior de lactentes com paralisia braquial perinatal (PBP). Trata-se de estudo experimental de caso único do tipo A-B com *follow-up*. Foram documentados semanalmente dados relativos à função do membro superior Escala de Movimento Ativo de dois lactentes, uma menina e um menino de 7 e 10 meses de idade, respectivamente. Após seis avaliações, três vezes por semana, durante duas semanas, na linha de base (A), a intervenção consistiu em aplicar FES Neurodyn® no músculo deltoide em combinação com treino de alcance de objetos, incentivando abdução e flexão anterior de ombro acima de 90°, durante seis semanas, totalizando 15 atendimentos (B). Os lactentes foram reavaliados duas vezes após um período de 15 e 30 dias sem intervenção (*follow-up*). A pontuação total obtida em cada dia de avaliação foi representada graficamente. Houve boa

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Part of this work was presented in a poster format in the Fifth Brazilian Congress of Neurofunctional Physiotherapy, that took place in October 11, 12 and 13, at Florianópolis, Santa Catarina.

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aceitação da eletroestimulação, sem nenhum efeito colateral. Ambos os lactentes apresentaram melhora da movimentação ativa do membro superior afetado após a intervenção e os ganhos foram mantidos no *follow-up*, com aumento de 9 e 7 pontos para os lactentes 1 e 2, respectivamente. Seis semanas de intervenção resultaram em mudanças individuais na função do membro superior dos lactentes, principalmente no aumento da amplitude de movimento ativo em flexão e abdução de ombro e flexão de cotovelo. Os resultados deste estudo sugerem o uso de estimulação elétrica como coadjuvante do treino do uso do membro superior afetado de lactentes com PBP.

Descritores | Neuropatias do Plexo Braquial; Estimulação Elétrica; Fisioterapia.

RESUMEN | El objetivo de este estudio fue evaluar el efecto del entrenamiento de alcance combinado con electroestimulación funcional (FES) sobre el movimiento activo de la extremidad superior de los lactantes con parálisis braquial perinatal (PBP). Este es un estudio experimental de caso único de tipo A-B con *follow-up*. Se registraron semanalmente datos sobre la función del miembro superior (escala de movimiento activo) de dos lactantes, una niña y un niño de 7 y 10 meses de edad, respectivamente. Después de seis evaluaciones, tres veces semanales, durante dos semanas, en la línea base (A), la intervención consistió en aplicar FES Neurodyn® en el músculo deltoides combinado con entrenamiento de alcance de objetos para estimular la abducción y la flexión anterior del hombro por encima de 90°, durante seis semanas, lo que totalizó 15 visitas (B). Se reevaluó a los lactantes dos veces tras el período de 15 y 30 días sin intervención (*follow-up*). El puntaje total que se obtenía en cada evaluación se representaba gráficamente. Hubo buena aceptación de la electroestimulación, sin efectos secundarios. Ambos lactantes tuvieron mejora en el movimiento activo de la extremidad superior afectada tras la intervención, y los beneficios se mantuvieron en el *follow-up*, con un aumento de 9 y 7 puntos para los lactantes 1 y 2, respectivamente. Seis semanas de intervención produjeron cambios individuales en la función del miembro superior de los lactantes, principalmente el aumento de la amplitud del movimiento activo en la flexión y abducción del hombro y la flexión del codo. Los resultados sugieren usar la electroestimulación como complemento en el entrenamiento de la extremidad superior afectada de lactantes con PBP.

Palabras clave | Neuropatías del Plexo Braquial; Estimulación Eléctrica; Fisioterapia.

INTRODUCTION

Perinatal brachial palsy (PBP) refers to flaccid paralysis of the upper limbs, secondary to a traumatic injury to one or more roots of the brachial plexus during the perinatal period[1]. The incidence of PBP ranges from 0.4 to 4.6 per 1,000 live births[2,3]. The most common type of PBP is Erb's palsy, which affects the roots of C5 and C6, and corresponds to 50-60% of cases[1]. PBP is generally transient, with partial or complete recovery of upper limb functions in the first three months of life, if neuropraxia has occurred[4]. However, in 10-30% of cases there is no spontaneous recovery. Therefore, the infant will have difficulties in using the affected upper limb, muscle weakness and may develop contractures and deformities[5,6]. In such cases, the lesion may have been of axonotmesis or neurotmesis type. The latter has a worse prognosis, and the infant will probably need surgical intervention[4].

Physiotherapy plays an essential role in stimulating the affected upper limb of infants with PBP in order to encourage active movement, decrease possible compensatory movements and increase muscle strength[7,8]. Studies suggest that strategies such as weight bearing and decubitus and positioning change could be adopted to improve muscle activity in infants with PBP[8,9]. Electrical stimulation is one of the resources that can be used to increase strength in plegic or paretic limbs, being widely used in children with cerebral palsy[10]. It is possible that the use of functional electrical stimulation (FES) may also optimize the rehabilitation process of children with PBP, as it stimulates sustained muscle contraction and recruits type II (i.e. fast-twitch) fibers[11].

The literature presents studies on electrical stimulation in adults with traumatic brachial plexus injury[12], with few studies in children. The only study found by the authors on the use of FES in infants before the second year of life was that of Berggren and Baker[13]. They reported the case of an infant with total PBP who was followed during the first two years of life. When he was between 11 and 14 months of

age, the infant was submitted to FES in elbow and wrist flexors and extensors, with the outcome of the improvement in the active movement of these joints[13]. Studies suggest that the younger the child, the greater the ability to recover the function of the upper limb affected by PBP[14]. However, it is not clear how conservative treatment can assist in this recovery process, as no studies were found that investigated the effect of training on the use of the affected upper limb in infants with this type of injury[15]. The aim of this study was to investigate the effect of FES combined with reach training on active movement of the affected upper limb in infants with PBP.

METHODOLOGY

Study design

A single-case AB experimental study with follow-up was carried out with phase A referring to the baseline (two weeks, evaluations three times a week), phase B referring to the intervention (six weeks, evaluations, and intervention three times per week). and the follow-up referring to two reassessments, 15 and 30 days after the end of the intervention (Chart 1). Those responsible for infants signed the Free and Informed Consent Form (ICF) before participating in the study.

Chart 1. Study design: distribution of assessments and days of intervention over the weeks

	Phase A: *Baseline*						Phase B: Intervention																		*Follow-up*			
Week	1			2			3			4			5			6			7			8			9	10	11	12
Evaluation	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X		X		X
Intervention							X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X	X				

Sample

The infants were selected at the physiotherapy school-clinic of the Faculdade Ciências Médicas de Minas Gerais (FCMMG). The study included infants aged between 4 and 12 months, with PBP of the Erb type (C5 and C6), group 1, according to the classification of Narakas[16] and axonotmesis-type lesion, according to the diagnosis given by the neurologist. The included infants were already included in a rehabilitation program when they started participating in the study.

Procedure

Initially, an interview was conducted with the family to collect data regarding chronological age, gestational age, weight, height, head circumference and Apgar score for each infant. The range of passive movement of the main muscles of the affected upper limb was evaluated to document possible muscle shortening. In addition, it was explained to the responsible for the infant that the use of electric current could cause a tingling sensation and that during the intervention, the infant's facial expressions and their interaction with therapists and objects would be monitored at all times, in order to identify possible discomfort, even if the infant did not demonstrate it through crying or compensations.

The infants were submitted to three weekly assessments of the active movement of the affected upper limb during phases A and B and two assessments corresponding to the follow-up. All evaluations were filmed so that, through video analysis, the active movement scale (AMS) was applied[17]. AMS is an instrument that rate the active movement of the affected upper limb of the infant and can be used in children from birth to adolescence. The score of 15 articular movements of the upper limb is graded on a scale ranging from zero to seven, totaling 105 points, based on the amount of movement observed within the available passive range of motion (ROM). The active movement performed with minimized gravity is scored from zero to four, while movements that overcome the action of gravity are scored from five to seven[17]. AMS was used to document the main outcome of interest in this study, which is included in the domain of body structure and function of the International Classification of Functionality, Disability and Health (ICF)[18]. It is a valid and reliable scale[17], capable of documenting the clinical evolution of the infant during the first year of life.

The evaluation also included a visual analysis of the quality of the movements performed with the affected limb in order to observe if the infant had compensatory movements, such as shoulder elevation, excessive scapular abduction and if he was able to keep the elbow close to the body, in addition to the postures and transfers that infants were able to make according to their age. This information was used to conduct the intervention in order to favor the active or active-assisted movement of the affected upper limb, according to what was expected for the infant's age. The evaluations were carried out by a physiotherapist with

more than 10 years of experience in evaluating infants with PBP. In turn, the intervention was carried out by academics who were already familiar with the infant and were responsible for the care during the internship period, under the direct supervision of a teacher.

The intervention consisted of the use of FES combined with encouraging active movement of the infant in visits of 50 minutes a day, three times a week, for six weeks. The equipment used was the six-channel digital FES Neurodyn®. Two self-adhesive spherical electrodes of 3 cm in diameter were fixed: the first in the middle deltoid muscle, just below the acromion, and the second in a lower position than the first, at a distance of approximately the size of the electrode. Initially, the frequency was adjusted to 10 pulses per second (pps) and the intensity was increased until it caused visible muscle contraction, in order to find the best positioning of the electrodes. Then, the intensity was decreased, and the stimulus frequency was adjusted to 30pps to achieve a smooth tetanic muscle contraction in shoulder abduction. The electric current used was symmetrical, with a pulse duration of 300 microseconds. The intensity (current amplitude in 1,000 amps) was individually adjusted according to the tolerance of each infant, varying between 15 and 30 thousand amps. The ON time was five seconds, with a two-second uphill ramp and a two-second lower ramp; the OFF time was 10 seconds, for 20 minutes. Whenever the current was passing, the infant was encouraged to reach some object[19]. Figure 1 illustrates the positioning of the electrodes and the intervention of the FES combined with encouraging the use of the affected upper limb (Figure 1).

Figure 1. Positioning of self-adhesive electrodes: the first attached to the deltoid muscle just below the acromion and the second at a distance of approximately 3cm from the first electrode

The infant treatment lasted 50 minutes, with 20 minutes of reach training combined with FES and another 30 minutes of reach training without electrotherapy, as the use of FES for more than 20 consecutive minutes could cause muscle fatigue in infants[20]. The details of the intervention are described in Chart 2.

Intervention	Duration	Objects to be reached	Position in which objects were placed	Postures in which the infant was encouraged to reach the objects
Range training combined with FES	20 minutes	Rings, trolleys, small balls, pieces of assembling or fitting blocks	Above the height of the infant's head in order to encourage him to pick up the object by performing anterior flexion or shoulder abduction above 90 °	Sitting Quadruped position Standing with support During transfers such as squatting and getting up, moving from lying to sitting and moving from sitting to quadruped position
Reach training without FES	30 minutes			

Statistical analysis

The sample was described in terms of age, sex, gestational age, weight, height, head circumference and Apgar. The total score obtained by each infant in the AMS on each evaluation day was plotted. A slope (celeration line) was calculated for the baseline phase to determine a developmental trend and its trajectory was extended to the other two phases. The difference between the baseline and intervention phases was determined by visual analysis of the proportion of points in the intervention phase that were above or below the trend line.

RESULTS

Two infants participated in this study, being the infant 1 female, seven months old, and infant 2 male, 10 months old. Infant 1 was born in vaginal delivery, with a gestational age of 40 weeks, birth weight of 3,650 g, height of 51 cm, head circumference of 36 cm and Apgar of 9 in the first and fifth minutes of life. In the initial evaluation, muscle shortening of the upper trapezius, biceps brachii, teres major, pectoralis major and pronators of the left forearm was documented. The affected upper limb was hypomobile and maintained in internal rotation and elevation of the shoulder, semiflexion of the elbow

and pronation of the forearm. The infant had limitations to perform weight bearing during transfers and difficulty in reaching, as this was done only when the object was placed close to his body. When reach required anterior shoulder flexion, the infant was unable to perform it. The manipulation of objects with an affected limb was difficult due to the infant's inability to perform forearm supination, resulting in negligence.

Infant 2 was born with vaginal delivery, with a gestational age of 40 weeks, birth weight of 4,775g, height of 53cm, head circumference of 38cm and Apgar score of 8 in the first minute and 9 in the fifth minute. In the initial evaluation, muscle shortening of the pectoralis major and teres major, elbow in semiflexion and hyperactivity of the upper trapezius was documented. The infant was able to reach objects up to half the range of motion of the anterior flexion and shoulder abduction against the force of gravity and was unable to actively supine the forearm.

The two infants who participated in the study tolerated electrical stimulation well and had no side effects, expression of discomfort or pain. Both were submitted to 15 intervention sessions during the six-week period, as there were three absences during that period due to the personal difficulties of those responsible. After the intervention, an improvement in the active movement of the affected upper limb in both infants was documented, assessed by means of AMS, mainly an increase in the range of active movement in shoulder flexion and abduction and elbow flexion. This improvement was maintained in the two reassessments performed 15 and 30 days after the end of the intervention. The gains were different for each infant, as the ability to perform active movement with the affected upper limb was different between them. Infant 1 was able to reach objects by performing anterior shoulder flexion against the force of gravity in less than half the range and had greater use of the affected upper limb compared to the beginning of the study. Infant 2, who was able to reach up to half the range of motion of the anterior flexion and shoulder abduction at the beginning of the study, was able to reach up to the end of the range of motion after the intervention. The two muscle groups that showed less changes after the intervention were the external shoulder rotators and forearm supinators, as the infants presented these movements only with the minimized action of gravity. Figure 2 shows the graphs of the total score obtained in the AMS of participants 1 and 2 over the entire study period.

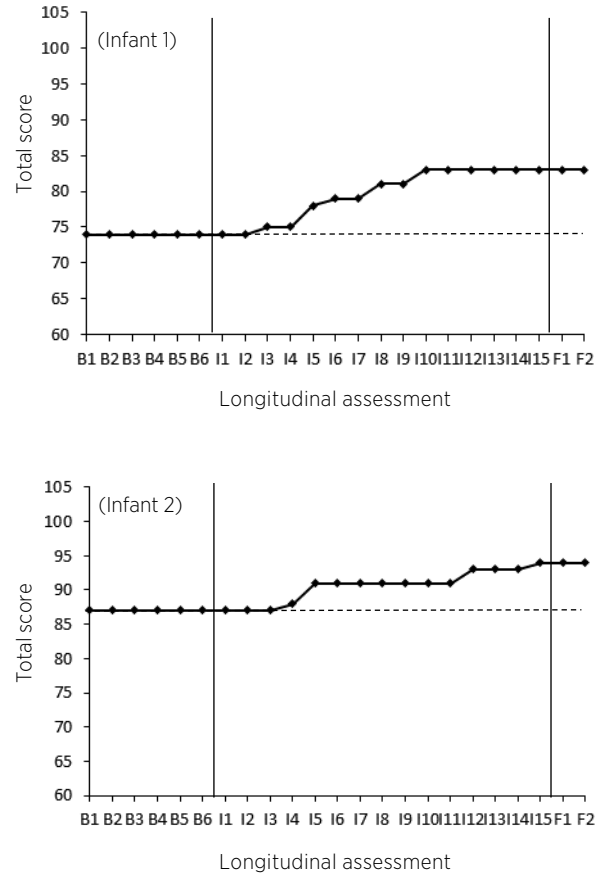

Figure 2. Graphs of the total score of the active movement scale (AMS) of infants 1 and 2 during the 23 longitudinal evaluations. The scores obtained in the baseline (B1-B6), intervention (I1-I15) and follow-up (F1e F2) are shown together with a line (.....), indicating the score obtained in the baseline phase, which was extended to the other two phases. This trend line is the celeration line

At the end of the range training intervention combined with the FES, both infants were able to maintain themselves on quadruped position with their arms extended, but infant 1 still needed support on the trunk to maintain stability and achieve reach in this posture. Improvement in the scapular stability of both infants was also observed, so that infant 2 was already able to bear weight on the affected limb and reach with the other upper limb.

DISCUSSION

This experimental single-case study documented the effect of combined reach training with FES on infants with PBP. The results showed that there was a good tolerance of infants to electrical stimulation, pointing out that this intervention can be a resource to enhance the use of the upper limb affected by PBP. As expected,

during the intervention period, infants were able to reach in a greater range than they were able to before the combined intervention with FES. The smallest change was documented in the external shoulder rotation and forearm supination movements, a result supported by the literature, which points out that these are the last muscle groups to present recovery of active movement in children with PBP[6].

This study proposed a conservative intervention for infants with PBP. The literature points to several studies on surgical intervention for infants with PBP who did not recover the function of the affected upper limb after the third or sixth month of life[8,21]. This study followed two infants who had already passed the age that the literature points out as the ideal period for recovery of the function of the upper limb in the case of damage to the roots of C5 and C6, which reinforces the effectiveness of the proposed intervention[8,15]. However, the comparison of these results with other studies that proposed conservative treatment in the first year of life is limited, as research is scarce. A randomized clinical trial investigated the effect of dosing an exercise program that included passive and active movement of the affected upper limb joints in infants with PBP. Although there was no difference between performing exercises once or three times a day, infants in both groups improved passive and active range of motion in reevaluations at 3, 6 and 12 months of age[22]. Another randomized clinical trial with older children, between three and five years old, showed that an intervention that included resistance exercises and weight bearing in the upper limb affected by PBP was not different from the same intervention combined with FES[19]. However, the recovery capacity in infants is higher in older children due to the neuronal recovery process that occurs in the first months after injury by PBP[3].

The rehabilitation of infants with PBP is very different from that of older children, as infants do not comply with verbal commands given by the therapist. For the infant to reach the objects, the intervention needs to be carried out in a playful context, with different toys, so that he is interested in reaching them. In this study, it was possible to identify, by the facial expression of the infants, that they were feeling something in their upper limb while the electric current was in the ON time, as the infant looked at the hand. Electrical stimulation drew the infant's attention to the affected upper limb, and this favored the use of that limb during reaching. In addition, infants did not cry or express difficulties during electrical stimulation. Therefore, a major contribution of this study is to show the feasibility of using FES in infants.

The literature points out that the use of FES is not indicated for many consecutive minutes due to the risk of causing muscle fatigue in infants[20]. Therefore, in this study, after reaching training with FES, the infant was encouraged to continue training without using FES. Physiotherapeutic care with infants is very dynamic. To keep them involved in the performance of outreach activities it was necessary to use different objects to be reached according to their interest. In addition, the infant actively changed his posture, for example, moving from sitting to quadruped position, and the therapist continued the reach training while interacting with him. Thus, the improvement in active movement that the infants presented can be attributed to the set of activities performed that had as main focus the reach training, which was carried out with and without the aid of electrical stimulation.

This study has some limitations. The two-week baseline period used, may not have been sufficient to establish a stable pre-intervention phase for the studied variables. However, it was not possible to use the baseline period equal to the intervention period, as there would be a risk of experimental attrition, that is, a threat to the internal validity of the study due to the maturation of the infants. The follow-up period was also short, which makes it impossible to affirm whether the gains obtained were maintained after the last assessment of the outcomes. Another limitation is small sampling, which limits the generalization of results. However, the experimental sigle-case design of this study allowed to document the changes in the active movement of each infant, under controlled conditions, considering their individual specificities. This study design is useful in health conditions such as PBP, as its prevalence after three months of age is low due to the spontaneous recovery that occurs in most cases.

CONCLUSION

The reach training combined with the FES increased the active movement of the affected upper limb of infants with PBP. There was good tolerance to the electric current and there were no complications during its use. Six weeks of intervention resulted in individual changes in the function of the upper limb of infants. The results of this study suggest the use of electrical stimulation as a training adjunct to the use of the affected upper limb

of infants with PBP. Further studies should be carried out to confirm the results obtained and to elucidate the benefits of electrotherapy in infants with PBP.

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