IMPACT OF EARLY PROSTHETIC FITTING ON PSYCHOMOTOR DEVELOPMENT OF CHILDREN WITH UNILATERAL CONGENITAL BELOW-ELBOW DEFICIENCY

VPLIV ZGODNJE OPREME S PROTEZO NA PSIHOMOTORIČNI RAZVOJ OTROK Z ENOSTRANSKO PRIROJENO PODLAKETNO AMPUTACIJO ZGORNJEGA UDA

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Izvleček

Uvod:

Cilj opreme s protezo in (re)habilitacije otroka po amputaciji je celostni funkcijski razvoj otroka. Primarna oprema otrok z enostransko prirojeno podlaketno amputacijo je estetska. Opremo s podlaketno protezo lahko izpeljemo v okviru ambulantne obravnave, običajno v starosti otroka od sedem do devet mesecev, pri čemer mora biti otrok vključen tudi v program fizioterapije. Cilj tako zgodnje opreme s protezo v zgodnjem psihomotoričnem razvoju je razvoj ustrezne predstave o telesu v možganih. S pomočjo proteze ne moremo povsem povrniti zmožnosti najrazličnejših vrst gibanja zapestja in roke, zmožnosti prilagajanja moči, koordinacije gibanja, spretnosti in natančnosti gibanja. Glede na to lahko otrok protezo uporablja skupaj z dominantno roko kot pomoč pri soročnih dejavnostih in stabilizaciji predmetov.

Metode:

Raziskava je bila izvedena na Kliničnem inštitutu za rehabilitacijo in ortopedske pripomočke v Zagrebu, v obdobju od leta 2012 do leta 2015. Vključenih je bilo 12 otrok (sedem deklet in pet fantov) z enostransko prirojeno podlaketno amputacijo roke, v starosti od tri do pet let, ki so bili opremljeni s prvo estetsko protezo v starosti od sedem do devet mesecev. Za oceno soročnih dejavnosti smo uporabili lestvico ABILHAND-Kids. Za oceno lastne telesne podobe smo uporabili vizualno analogno lestvico (VAS).

Abstract

Background:

The aim of prosthetic fitting and associated (re)habilitation is promotion of functional, psychological, cognitive and social development with the prosthesis as a new biomechanical entity. The primary prosthesis for children with unilateral congenital below-elbow deficiency (UCBED) is aesthetic. It is applied in outpatient setting, usually at the age of 7-9 months, along with developmental physiotherapy. The objective of such an early prosthetic fitting is the timely creation of engrams through early psychomotor development. The large variety of wrist and hand movements with all modes of power, coordination, agility and precision of movement cannot be fully restituted with the prosthesis. Hence, the prosthesis is utilised along with the dominant hand as an aid for fixation and synergy.

Methods:

A survey was conducted at the Clinical Institute for Rehabilitation and Orthopedic Aids in Zagreb in the period from 2012 to 2015 on 12 children (7 girls and 5 boys) with UCEBED aged 3-5 years, primarily fitted with pasive prosthesis at the age of 7-9 months. Functionality in bimanual activities was tested using the ABILHAND-Kids scale. Self-assessment of body image was performed using a reverse visual analogue scale (VAS).

Results:

The raw ABILHAND-Kids scores were statistically significantly

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Resultati:
Rezultati ocene z lestvico ABILHAND-Kids so bili v povprečju statistično značilno nižji pri izvedbi nalog s protezo (povprečni rezultat 26,1) v primerjavi z izvedbo nalog brez proteze (povprečni rezultat 30,9; $p < 0,001$). Hkrati pa smo ugotovili, da je bilo zadovoljstvo s telesno podobo, povezano z družbeno sprejemljivostjo in pasivno uporabo proteze v vsakdanjem življenju v povprečju statistično značilno višje, kadar so otroci uporabljali protezo, kot takrat, ko je niso uporabljali (povprečna VAS oceno 9,8 oziroma 3,7; $p < 0,001$).

Zaključek:
Celovita (re)habilitacija otrok s protezo za zgornji ud zahteva celostni individualni pristop, ki je usmerjen v otrokove težave in se odvija v okviru strokovne skupine. Takšna (re) habilitacija je potrebna, da bi otroku lahko omogočili svobodo gibanja in neomejeno igro, izražanje njegove lastne osebnosti, potreb, želja in sposobnosti. Zgodnja oprema s protezo, stalno usposabljanje za opravljanje dejavnosti, ki so primerne za določeno starost, vključevanje staršev in stalno nošenje proteze so najpomembnejši dejavniki, ki vplivajo na otrokovo sprejemanje estetske proteze in ki imajo velik vpliv na otrokov normalni psihološki, socialni in gibalni razvoj ter rast otroka.

Ključne besede:
okvara zgornjega uda; otrok; proteza; (re)habilitacija

Results:
Results of the ABILHAND-Kids questionnaire were statistically significantly lower on average when performing functional tasks with the prosthesis as compared to performing them without the prosthesis (mean score 26.1 vs. 30.9, $p < 0.001$). However, satisfaction with body image related to social acceptance and passive application of prosthesis in everyday life was statistically significantly higher on average with the prosthesis than without it (mean VAS rating 9.8 vs. 3.7, $p < 0.001$).

Conclusion:
The comprehensive habilitation of children with a prosthetic limb requires an individual, holistic and team-based problem-oriented approach. It is necessary to enable the child's freedom of movement and unrestricted play, along with his/her expression of personality, needs, desires and abilities. Early prosthetic fitting, continuous training for carrying out activities appropriate for the given age, involvement of parents and permanent wearing of the prosthesis are the most important factors for the acceptance of pasive prosthesis, which has a significant impact on the normal psychological, social and motor development and growth of the child.

Key words:
upper limb impairment; child; prosthesis; (re)habilitation

INTRODUCTION

Congenital deficiency is a lack of part or whole limb evident at birth. Congenital aplasia of upper extremities is rare; the most common is the congenital aplasia of the hand with an incidence of 1 in 20.000 new-borns. The incidence of forearm hemimelia in terms of aplasia or hypoplasia of the radius, radial deficiency or congenital varus of the hand is 1 in 100.000 new-borns (1).

Early intervention and support are critical to the family of a child with unilateral congenital below-elbow deficiency (UCBED). Every child is unique and has got to, together with his/her family, become as early as possible a part of a comprehensive rehabilitation team consisting of an orthopaedic surgeon, PRM specialist, physical therapist, occupational therapist and prosthetist within a specialised medical institution. Many clinicians encourage early prosthetic fitting as a basis for normal neurodevelopment of children with UCBED and secondarily, as a means of prevention of overuse syndrome later in life (2).
The goal of habilitation and prosthetic fitting is to encourage functional, psychological, cognitive and social development with the prosthesis as a new biomechanical entity. The first prosthesis for UCBED is pasive and is applied in outpatient setting usually at the age of seven to nine months in the course of developmental physiotherapy. The objective of such an early prosthetic fitting is the timely creation of engrams during the most adaptable phase of psychomotor development. Physiotherapy and occupational therapy commence, if indicated, during the first prosthetic fitting by using the prosthesis in activities such as retrieval and capture of object, rolling, crawling, sitting and getting up, postural adjustment and strategy changes. Follow-up and (re)habilitation are carried out once a year or as needed, depending on the developmental stage of the child and. The use of the prosthesis during activities reduces compensations, musculature develops symmetrically and negative effects on postural changes of the spine are minimised (2).

The huge variety of fist and hand movements with all the modes of power, coordination, agility and precision of movement cannot be restituted with the prosthesis. The prosthesis is therefore utilised along with the dominant hand as an aid for fixation and synergy.

Children with UCBED usually do not need pre-prosthetic habilitation because these are healthy, with no associated illnesses or

impairments. Appropriate prosthetic fitting, continuous habilitation, family engagement and constant interest in the therapy are key factors for the development of child's independence. However, it is necessary to separate the concept of functionality with the prosthesis from merely wearing it (3).

METHODS

A survey was conducted at the Clinical Institute for Rehabilitation and Orthopedic Aids of the University Hospital Centre in Zagreb in the period from 2012 to 2015, comprising 12 children (seven girls and five boys) with UCEBED aged three to five years, primarily fitted with an pasive prosthesis at the age of seven to nine months. Functionality in age-dependent bimanual activities was tested using the ABILHAND-Kids scale (ABILHAND-Kids, i.e., Manual Ability Measure for Children with Upper Limb Impairments) (5). Satisfaction with body image (as related to social acceptance and passive application of prosthesis in everyday life) was assessed using a reverse visual analogue scale (VAS) adapted to the children's age (14).

Manual ability, as measured by the ABILHAND scale and its version for children is defined as the capacity to manage daily activities that require the use of the upper limbs, whatever the strategies involved (6). It refers to the Activity domain of the International Classification of Functioning, Disability and Health (ICF) (13). The ABILHAND-Kids was developed in France. It assesses the child's ability to perform everyday manual activities by means of a parent-report questionnaire that assesses the child's difficulty in performance as perceived by his/her parents. The 21 assessed activities are mostly bimanual tasks. Each task is rated on a 3-point degree-of-difficulty scale. The ABILHAND-Kids was developed using the Rasch measurement model, so that it is a linear measure with a unidimensional scale. The authors note that it takes about 10 minutes to complete (4, 5). Its characteristics can be summarised as follows (6):

- Each item is answered on a 3-level scale (impossible, difficult, easy).
- The item difficulty increases with bimanual involvement.
- Measurement range is approximately 9 logits.
- Measurement error is 0.42 logit in the centre of the scale.
- The smallest measurable difference is 0.19 logit in the centre of the scale.
- Separation reliability was 0.94 in a sample of 113 children with cerebral palsy.
- Test-retest reliability was 0.91 after a delay of 25±13 days.

Visual Analogue Scale (VAS) is a type of psychometric response scale. It is a measurement instrument for subjective characteristics or phenomena that cannot be directly measured. When responding to a VAS item, respondents specify their level of agreement to a statement by indicating a position along a continuous line between two end-points (14).

The data were statistically analysed using paired *t*-test and Pearson correlation. The analyses were performed using Microsoft® Excel 2010 software.

RESULTS

All the children from study are able to incorporate the prosthesis into their movement strategies and postural adjustment. Most children will choose to do a one-handed task with the intact upper limb just as a person with two healthy hands will prefer his/her dominant hand to complete a one-handed task. According to the parents' ratings, children are more independent in bimanual activities without the prosthesis, because they are fitted with a passive, aesthetic prosthesis; this goes especially for the following activities: sharpening a pencil, zipping up trousers, buttoning up trousers, opening the cap of a toothpaste tube, squeezing toothpaste onto a toothbrush and fastening the snap of a jacket. Three-year old children cannot perform most of those activities alone. Of the twenty-one tasks listed in the ABILHAND-KIDS for three to five-year-olds, unscrewing a bottle cap and opening a jar of jam was perceived by the parents to be the most difficult both with and without the prosthesis. Children do not use their prosthesis when they filling a glass with water. Taking off a T-shirt and switching on a bedside lamp are the simplest activities to perform with the prosthesis. Children achieve some degree of independence regardless of whether they wear a prosthesis or not. Use of a prosthesis by children with UCBED is not associated with relevant differences in function in daily-life activities. Children with unilateral congenital below-the-elbow deficiency have nearly normal, but still not normal, function, and the presently available prostheses cannot compensate for that.

The raw ABILHAND-Kids scores obtained using and not using the prosthesis are presented in Figure 1. The children are listed in increasing order of the score obtained using the prosthesis. The average score without the prosthesis (mean 30.9, SD 8.8) was statistically significantly higher than with the prosthesis (mean 26.1, SD 8.4; $p = 0.0003$ from paired *t*-test).

Children with UCBED perceive themselves as different, not necessarily bad, but unique. They are very proud of their prosthesis

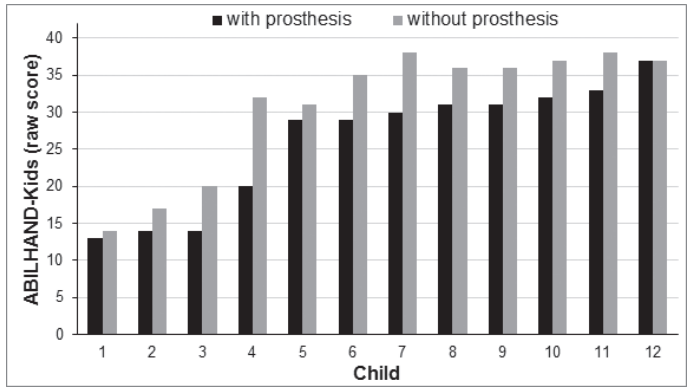


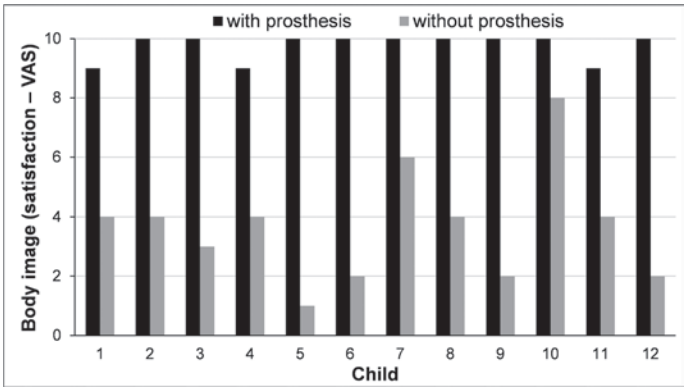
Figure 1: *Raw ABILHAND-Kids scores obtained with and without the prosthesis. (The children are listed in increasing order of the score obtained using the prosthesis.)*

and they often do not want to go to the children's nursery without their prosthesis.

The VAS readings referring to using and not using the prosthesis are presented in Figure 2. The children are listed in the same order as in Figure 1. The average score with the prosthesis (mean 9.8, SD 0.5) was statistically significantly higher than without the prosthesis (mean 3.7, SD 1.9; $p < 0.0001$ from paired t-test).

Figure 2: *VAS readings referring to using and not using the prosthesis. (The children are listed in the same order as in Figure 1.)*

The two ABILHAND-Kids scores were highly positively correlated among themselves ($r = 0.93$, $p < 0.0001$); the two VAS readings were not correlated ($r = 0.10$, $p = 0.7458$). The difference between



the score with the prosthesis and the score without the prosthesis on the ABILHAND-Kids was not statistically significantly correlated with the difference between the two VAS readings ($r = -0.35$, $p = 0.2659$).

DISCUSSION

On average, the scores on the ABILHAND-Kids scale were statistically significantly higher when the children did not use the prosthesis than when they did, thus indicating decreased manual ability when using the prosthesis (i.e., the parents perceiving the child as having more difficulties in performing the required tasks with the prosthesis than without it). Conversely, the prosthesis improved the child's perceived body image in the sense of higher level of satisfaction with social acceptance and passive application of prosthesis in everyday life. The absence of a statistically significant correlation between the effect of the prosthesis on the manual ability and the effect of the prosthesis on satisfaction with body image further indicates that these two aspects are not necessarily related.

Some therapists believe that earlier fitting results in better incorporation of the prosthesis into the child's body image and that early fitting may be essential for normal neuromuscular development (7, 8). There has been little research on prosthetic use by children with a unilateral congenital below-the-elbow deficiency, and there are no established guidelines regarding the prescription of prostheses or methods for assessing use and function by these children (10). Early prosthetic fitting provides the opportunity to develop bimanual skills, useful prehensile activities, along with incorporating the prosthesis into the body image. Early fitting may ultimately contribute to better prosthetic tolerance and wear patterns and may prevent an asymmetrical posture and spinal curvature (9). Prostheses may help with social acceptance or may be useful as tools for specialized activities, but they do not appear to improve function or quality of life, which are nearly normal for children with unilateral congenital below-the-elbow deficiency regardless of whether they wear a prosthesis (10). Even if a prosthesis does not meet all of the child's needs and if its use is not continued, a prosthesis may serve a short-term purpose or benefit during a certain developmental period (11). The first fitting is a passive device, which enables the infant to oppose the sound hand for gross two-handed grasping and develops a prosthetic wearing pattern that will ease the transition into an activated limb (11).

In addition to the small sample size, the main limitation of our study is that we used raw ABILHAND-Kids scores rather than the truly linear logits. However, the conversion is not available for the population that we studied, while our sample is much too small to conduct our own Rasch analysis. A possibility might have been to use the online conversion tool for children with cerebral palsy (12).

CONCLUSION

The complex habilitation of children with a prosthetic limb requires an individual, holistic and team problem-oriented approach. It is necessary to enable the child freedom of movement and unrestricted play, and the expression of personality, needs, desires and abilities. Early prosthetic fitting, continuous training for carrying out activities related to the age, involvement of parents and permanent wearing of the prosthesis are the most important factors for the acceptance of cosmetic prosthesis that has a significant impact on the normal psychological, social and motor development and growth of the child.

Prostheses have been assumed to improve the function of children with unilateral deficiency, but the pasive one did not do so in this study. Children with unilateral congenital below-elbow deficiency have close to normal but not normal function and current prostheses are not normalizing function. In this study the average ABILHAND-KIDS score without the prosthesis was statistically significantly higher than with the prosthesis which means that children achieve some degree of independence regardless of whether they wear a prosthesis or not. Early prosthetic fitting provides the opportunity of incorporating the prosthesis into the body image and may help with social acceptance. The prosthesis improved the child's perceived body image in the sense of higher level of satisfaction with social acceptance and passive application of prosthesis in everyday life. A further study should be undertaken to assess long-term impact of early prosthetic fitting on psychomotor development of children with UCBED.

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