SPEECH IMPROVEMENT IN CHILDREN WITH CEREBRAL PALSY BY "BRAIN–COMPUTER–HAND EXOSKELETON" NEUROINTERFACE REHABILITATION

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As explained earlier, neurorehabilitation sessions involving the use of the non-invasive “brain – computer – hand exoskeleton” interface reduce hand muscle spasticity and improve motor skills in children with cerebral palsy (CP). However, the changes in the patients’ speech functions and their relationship with the upper limb mobility have not been analyzed. The study was aimed to assess the correlation between the motor and speech functions of children with CP, as well as to detect the changes in motor realization of speech production following complex treatment of patients including sessions of neurorehabilitation. The study involved children with CP aged 6–15. The index group (n = 40, 16 girls, 24 boys) received complex resort treatment with the course of neurorehabilitation, while the comparison group (n = 20, 10 girls, 10 boys) received standard resort treatment. A significant (p < 0.001) correlation between the total ABILHAND-Kids score and the indicators of speech production motor realization was revealed. In patients of the index group, complex treatment with the course of neurorehabilitation resulted in the significant (p < 0.001) decrease in hand spasticity and the increase in the total ABILHAND-Kids score and speech scores. No significant changes of these indicators were revealed in children of the comparison group. Beneficial effects of neurorehabilitation may be based on the enhanced plasticity of the neural circuits responsible for planning and execution of complex hand movements, as well as speech processes. The findings can be used to develop new methods for correction of motor and cognitive spheres in children with CP.

Keywords: children, cerebral palsy, speech, brain–computer interface

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УЛУЧШЕНИЕ РЕЧИ У ДЕТЕЙ С ДЦП НА ФОНЕ РЕАБИЛИТАЦИИ С ПРИМЕНЕНИЕМ НЕЙРОИНТЕРФЕЙСА "МОЗГ–КОМПЬЮТЕР–ЭКЗОСКЕЛЕТ КИСТИ"

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Как было показано ранее, сеансы нейрореабилитации с применением неинвазивного интерфейса «мозг–компьютер–экзоскелет кисти» снижают у детей с детским церебральным параличом (ДЦП) спастичность мышц кисти и улучшают двигательные навыки. Однако изменение речевых функций пациентов и их связь с подвижностью верхних конечностей не анализировали. Целью исследования было проанализировать связь между двигательными и речевыми функциями детей с ДЦП, а также выявить изменения моторной реализации высказывания у пациентов в результате комплексного лечения, включающего сеансы нейрореабилитации. В исследовании приняли участие дети с ДЦП в возрасте 6–15 лет. Основная группа (n = 40, 16 девочек, 24 мальчика) проходила комплексное санаторно-курортное лечение с курсом нейрореабилитации, а группа сравнения (n = 20, 10 девочек, 10 мальчиков) — стандартное санаторно-курортное лечение. Выявлена статистически значимая (p < 0,001) взаимосвязь между величиной суммарного показателя шкалы «Возможности кисти — дети» («ABILHANDKids») и показателями моторной реализации высказывания. Комплексная терапия с курсом нейрореабилитации привела у пациентов основной группы к статистически значимым изменениям (p < 0,001): снижению спастичности кистей рук, росту суммарного показателя «ABILHANDKids» и показателей речи. У детей группы сравнения статистически значимых изменений данных показателей не выявлено. Основой позитивных эффектов нейрореабилитации может быть усиление пластичности нейронных цепей, контролирующих выполнение сложных движений рук, а также речевые процессы. Полученные данные могут быть использованы при разработке новых методов коррекции двигательной и когнитивной сферы детей с ДЦП.

Ключевые слова: дети, церебральный паралич, речь, интерфейс мозг–компьютер

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Today it is acknowledged that cerebral palsy (CP) is much more than "postural and motor impairment", it is often associated with a broad spectrum of dysfunctions that includes cognitive, language, and sensory perception disorders [1]. As stated in the recently published review, 30–87% of patients with CP have speech disorders [2]. A more severe motor impairment is associated with the more severe speech disorders [1, 3–5]. The correlation analysis has made it possible to reveal the relationship between the severity of motor impairment and the severity of speech disorder in patients with CP aged 10–12 [6].

Similarity of motor and speech disorders associated with CP is explained by anatomical proximity of the cortical speech and motor regions, as well as their pathways. Functional similarity of the speech and motor systems has been also noted: organization of each speech act and movement requires preserved explicit kinesthetic perception that goes along with any movement of articulation and other muscles [2]. That is why the exercises designed to overcome motor deficits (especially in hands) are recommended to develop speech functions in children.

Currently, of particular relevance are methods for rehabilitation of the limb motor functions in patients with CP based on the use of non-invasive brain-computer interfaces (BCI) and the principles of biofeedback. Such methods make it possible to reinforce innate physiological resources of the child's brain [7]. Originally, the number of studies showed the potential for using BCI in children with CP in order to detect when the patient imagines limb movement or movement intention based on the analysis of EEG dynamics [8–10]. The children used BCI to control the cursor movement or various game objects on the computer screen. The researchers from South Korea, who used the results of these and some other studies, utilized a BCI integrated with the electrical stimulator of the wrist extensor muscles [11]. Electrical stimulation was launched based on the on-line analysis of EEG parameters when the patient imagined hand extension. Children with CP showed improvement of the parameters of hand movement execution and focus after a series of sessions.

Later the non-invasive "brain–computer–hand exoskeleton" interfaces that identified typical EEG patterns associated with kinesthetic motor imagery and triggered movement of the exoskeleton "gloves" were used for rehabilitation of children with CP [12, 13]. It has been found that the neurorehabilitation sessions involving the use of such non-invasive BCIs improve the effectiveness of rehabilitation measures applied during the resort phase of treatment. As a result, hand spasticity is reduced, muscle strength and the range of hand motion are improved, and the range of everyday skills is expanded. This promotes socialization of patients with CP. However, the changes in speech functions of patients have not been analyzed in the above studies. That is why our study was aimed to assess the relationship between the motor and speech functions of children with CP and to detect the changes in the patients' motor realization of speech production after the complex resort treatment including sessions of neurorehabilitation.

METHODS

Sample characteristics

The study was performed at the Health and Rehabilitation Technology Centre (V.I. Vernadsky Crimean Federal University) and the Gelilovitch "Chaika" Sanatorium for Children and their Parents for children with neurological disorders (Republic of Crimea). The study involved 60 children aged 6–15, who underwent health resort rehabilitation. Inclusion criteria: the diagnosis of CP according to ICD-10; hemiparesis with the motor activity level not exceeding III according to the Gross Motor Function Classification System for Cerebral Palsy (GMFCS) in the patient's structure of neurological disorder. Exclusion criteria: refusal to participate in the study obtained from the patient, his/her parent or legal representative; motor activity level exceeding III according to GMFCS; aphasia; epilepsy not responding to medication; vision problems not allowing the patient to see the instructions on the screen; moderate, severe or profound mental retardation (F71–73 according to ICD-10).

The index group included 40 individuals (16 girls, 24 boys) aged 6–15, who underwent complex resort treatment including the course of neurorehabilitation with the use of the Exohand-2 "brain–computer–hand exoskeleton" interface. The comparison (control) group included 20 children (10 girls, 10 boys) aged 7–13, who underwent complex resort treatment involving the use of standard methods. It should be noted that parents more often registered their children for the course of neurorehabilitation when the children had more severe motor function impairment. Thus, no randomization when forming the groups of patients is a limitation of our study. The complex resort treatment received by both groups of patients included the following: exercise therapy, paretic muscle massage, pelotherapy, hydrokinesitherapy in thermal mineral water, electrical stimulation of muscles that were antagonists of paretic ones.

Assessment of motor and speech function parameters

The following scales were used to assess the upper limb motion range:

1. Modified Ashworth Scale (MAS) allowing the neurologist to estimate spasticity by assessing the resistance experienced during passive range of motion using the 5-point scale (0–4).
2. ABILHAND-Kids, the questionnaire allowing the parents to assess the child's upper limb motor function when performing daily activities (3 levels of the ability to use the skill: "Impossible", "Difficult" or "Easy"). This scale showed responsiveness and high sensitivity when used to detect changes after the intense training of children with CP. It was recommended for monitoring of the patients' functional state in clinical trials [14]. In addition to standard action performance indicators (impossible to perform – X0, difficult to perform – X1, easy to perform – X2), a cumulative indicator (X) ranging between 0 and 42, which was calculated according to the formula X = X1+ 2X2, was used to assess functions in general [12].

Neuropsychological diagnosis of speech disorder in children was performed in accordance with the guidelines [15]. The level of motor realization of speech production was assessed by three methods.

1. Assessment of oral praxis and articulatory motility (evaluation of lip and tongue movement, pouting stretch according to the instructions or following the model). The maximum score was 30.
2. Assessment of pronunciation of sounds (repeating words). The maximum score was 30.
3. Making sentences based on the pictures (the child was presented a series of pictures, such as "a boy washes his hands", which he/she had to describe using one sentence). Correctness of word order, agrammatism and paragrammatic errors were taken into account during the test. The maximum score was 45.

Table 1. Hand spasticity scores according to the Ashworth scale in patients of the index and comparison groups before and after treatment (Me [Q_1; Q_3])

Spasticity (score)	Before treatment	After treatment	*p*
Index group, *n* = 40			
Left hand	2,0 [1,5; 2,0]	1,0 [1,0; 2,0]	*p* < 0,001
Right hand	2,0 [2,0; 3,0]	2,0 [1,0; 2,0]	*p* < 0,001
Comparison group, *n* = 20			
Left hand	2,0 [1,0; 3,0]	1,0 [1,0; 2,0]	*p* = 0,028
Right hand	2,5 [1,0; 3,0]	2,0 [1,0; 2,5]	*p* = 0,018

Rehabilitation procedures

Each child in the index group had 10 sessions of rehabilitation procedures involving the use of the complex consisting of non-invasive BCI and Exohand-2 hand exoskeleton (manufactured by Exoplast LLC, Moscow, in accordance with RU No. RZN 2018/7681). The non-invasive BCI operation is based on the analysis of EEG patterns emerging when an individual imagines hand extension. The program ensures detection of kinesthetic motor imagery based on the EEG pattern analysis, generates the visual feedback signal and issues commands to control the hand exoskeleton.

During neurorehabilitation training the patient was seated in a chair in front of the computer monitor, on which visual instructions were displayed. Hands were housed inside the exoskeleton “gloves”. There was a white round mark for gaze fixation in the center of the screen with three arrows around it, which changed color to mark the instructions. The patient executed the following commands: to relax, to kinesthetically imagine the left or right hand extension. To produce a particular kinesthetic image when imagining movement, the children were instructed as follows: “Imagine you have a small ball in your hand, you open your hand and drop it. Feel this movement”. When the patient executed the task precisely, the mark for gaze fixation changed its color to green (the color intensity was dependent on EEG parameters), the exoskeleton executed the appropriate movement, and the hand was passively extended. Thus, the combined visual and kinesthetic feedback signal was generated.

Initial assessment of the upper limb motion range and speech function parameters in children of the index and control groups was performed on day two after admission to the sanatorium. The index group patients took a course of neurorehabilitation consisting of 10 sessions (every day except Sunday) starting from day three of resort treatment according to the same scheme: three times per session of 8 min with a rest break of at least 5 min. The motor imagery task for each hand was repeated 24 times during the session. The percentage of correct answers of the classifier (which triggered the exoskeleton and ensured passive extension of the hand) during the first and second session was about 60%, while after the patient’s training, by the end of the course, it reached 75–80%. On the next day after the end of the course (day 14–15 of stay in the sanatorium) the data on motor and speech activity of the index group patients were acquired again. Parameters of controls were also assessed on day 14–15 of resort treatment.

Other details of the method have been reported earlier [12, 16].

Statistical analysis

Statistical data processing was performed using the Statistica 12 software (StatSoft Inc.; USA). The distribution of the studied indicators was assessed using the Shapiro–Wilk test. When the distribution was normal, the data were presented as mean and standard error of the mean; the Student's t-test was used to assess intergroup differences. When the distribution was non-normal, statistical data were presented as median and interquartile range (Me [Q_1; Q_3]); the Mann–Whitney *U* test was used to assess the intergroup differences, while the intragroup differences were assessed using the Wilcoxon signed rank test. Sperman’s rank correlation was used to calculate the correlation coefficients. The differences and correlation coefficients were considered significant at $p < 0.05$.

RESULTS

Characteristics of motor and speech functions before rehabilitation procedures

The average age of children in the index and control groups was 10.2 ± 0.4 and 10.1 ± 0.3 years, respectively, there were no significant differences ($p = 0.92$). Among patients of the index group, left-sided hemiparesis was revealed in 11 individuals, and right-sided hemiparesis was found in 29 study

Table 2. Everyday activity management scores according to the ABILHAND-Kids scale in patients of the index and comparison groups before and after treatment (Me [Q_1; Q_3])

Everyday activity management (score)	Before treatment	After treatment	*p*
Index group, *n* = 40			
Impossible	1,5 [0,0; 7,0]	0,0 [0,0; 6,0]	*p* = 0,001
Difficult	8,0 [3,5; 14,0]	7,0 [3,0; 13,0]	*p* = 0,055
Easy	6,0 [0,0; 12,0]	7,0 [0,0; 16,0]	*p* < 0,001
Total score	23,0 [14,0; 33,0]	27,0 [16,0; 37,0]	*p* < 0,001
Control group, *n* = 20			
Impossible	2,0 [0,5; 4,5]	2,0 [0,0; 3,0]	*p* = 0,068
Difficult	9,0 [6,0; 13,0]	8,0 [6,0; 13,5]	*p* = 0,593
Easy	7,0 [4,0; 12,0]	7,0 [4,0; 12,0]	*p* = 0,593
Total score	27,5 [13,5; 33,0]	27,5 [15,5; 32,5]	*p* = 0,593

Table 3. Motor realization of speech production in patients of the index and comparison groups before and after treatment (Me [Q_1; Q_3])

Spasticity (score)	Before treatment	After treatment	*p*
Index group, *n* = 40			
Oral praxis and articulation	20,0 [13,0; 25,0]	24,0 [16,0; 27,0]	$p < 0{,}001$
Pronunciation of sounds	18,0 [12,0; 26,0]	20,0 [12,0; 28,0]	$p < 0{,}001$
Sentence making	22,0 [5,0; 35,0]	25,0 [6,0; 40,0]	$p < 0{,}001$
Comparison group, n = 20			
Oral praxis and articulation	23,0 [14,0; 27,0]	23,0 [15,5; 26,0]	$p = 0{,}800$
Pronunciation of sounds	21,5 [12,0; 25,0]	21,5 [13,0; 25,0]	$p = 0{,}109$
Sentence making	24,0 [18,5; 36,0]	25,5 [19,0; 37,0]	$p = 0{,}237$

participants. In the comparison group (controls), left-sided hemiparesis was revealed in seven children, and 13 children had right-sided hemiparesis. Among patients with left-sided hemiparesis the total ABILHAND-Kids score was 27 [19; 34], while children with right-sided hemiparesis had a slightly lower score, 23 [12; 32]. However, the differences in this parameter between patients with left-sided and right-sided hemiparesis were non-significant ($p = 0.27$). The patients with right-sided hemiparesis also had lower scores for pronunciation of sounds, oral praxis and sentence making than children with left-sided motor impairment, however, the differences were non-significant ($p = 0.18–0.93$).

The hand spasticity scores of patients of the index and comparison groups are provided in Table 1, the ABILHAND-Kids scores for daily activity management are presented in Table 2, and the characteristics of motor realization of speech production are provided in Table 3. There were no significant differences between the scores of the groups before treatment.

Calculation of the Spearman's rank correlation coefficients showed that the left hand spasticity scores determined in all the surveyed children ($n = 60$) before treatment negatively correlated with the scores for oral praxis, articulatory motility, pronunciation of sounds, making sentences from pictures ($r = –0.37, –0.36, –0.30$ at $p = 0.004, 0.005$ and 0.019, respectively). The right hand spasticity scores correlated significantly with the scores for pronunciation of sounds ($r = –0.49$ at $p < 0.001$), i.e. the higher the level of hand spasticity, the lower the level of speech production motor realization. The total ABILHAND-Kids score and the scores for oral praxis, pronunciation of sounds and making sentences from pictures shows a significant ($p < 0.001$ in all cases) positive correlation ($r = 0.69, 0.62, 0.62$, respectively), i.e. the better preserved the upper limb function, the higher the level of speech production motor realization. An example of such correlation between the score for hand function and the ability to adequately pronounce sounds is provided in Fig.

Characteristics of motor and speech functions after rehabilitation procedures

A significant decrease in the left and right hand spasticity after both standard resort treatment and complex treatment including the course of neurorehabilitation was revealed in both groups of children (Table 1). However, the analysis of daily activity management based on the parent-reported data (total ABILHAND-Kids score) made it possible to reveal a significant improvement in the index group only (Table 2). The significant improvement of the speech production motor realization level

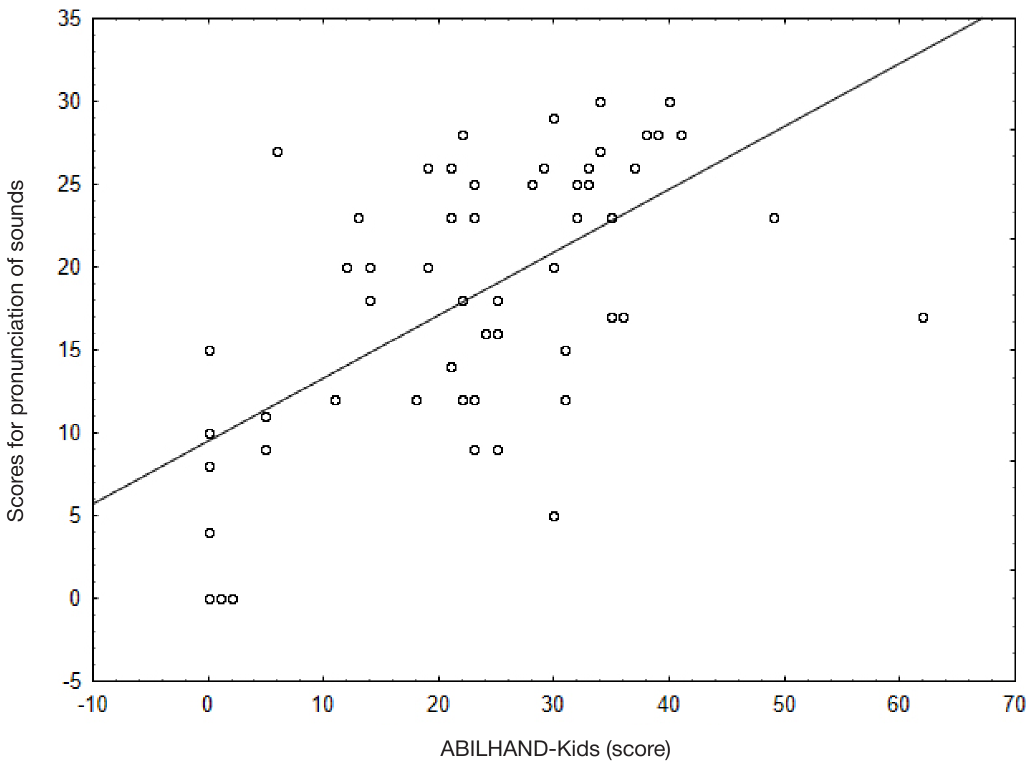

Fig. Correlation between the ABILHAND-Kids scores and scores for pronunciation of sounds in 60 children with CP was also found only in the index group children (Table 3). Oral praxis, pronunciation of sounds and making sentences from pictures are the components of speech ability and can be considered as the repeated measurements requiring the use of Bonferroni correction for four comparisons. However, significance of differences is retained after applying the correction (as can be seen from p-values).

It should be noted that no significant differences in the studied indicators have been found after treatment. This may be due to the fact that children with the slightly lower scores for everyday skills and motor realization of speech production were included in the index group, where the more complex treatment including the course of neurorehabilitation was used, because of their parents' desire. Their scores did not significantly exceed that of the comparison group even after treatment.

Meanwhile, the following facts further indicate the efficiency of complex resort treatment that involves rehabilitation procedures including the course of neurorehabilitation. In the index group consisting of 40 children, oral praxis and articulation improved in 33 individuals (83%), pronunciation of sounds improved in 24 (60 %), and sentence making improved in 31 patients (78%). Among 20 children of the comparison group who received standard resort treatment only, improvement of scores for oral praxis and articulation associated with pronunciation of sounds was found only in three individuals (15%), while the sentence making improvement was revealed in four patients (20%).

DISCUSSION

We have gathered evidence suggestive of the correlation between the upper limb function parameters and the level of speech production motor realization. The findings confirm the theory that actions including the sequence of fine motor skill components and actions that ensure speech production involve the same cognitive and motor neural network [3]. It has been shown that hand function improvement following a series of neurorehabilitation sessions in children with CP is associated with the increase of scores for oral praxis, articulation, pronunciation of sounds and making sentences from pictures. Such combination of changes may me based on the plasticity processes enhancement not only in the neural circuits of motor and sensorimotor areas of the neocortex responsible for planning and execution of complex hand movements, but also in adjacent conventional speech areas (for example, Broca's area) showing increased activity during execution of sequential upper limb movements [17]. It is interesting to note that improvement of fine motor skills in healthy subjects reported in the recent experiments on teaching stone-tool making also resulted in the development of neural centers and paths involved in generation of speech [18, 19].

One of the factors of enhanced nervous tissue plasticity can be rearrangement of the synthesis and binding of neurotrophic factors secreted mainly by neurons and glia. It has been shown that successful rehabilitation of children with CP involving the use of the "brain–computer–hand exoskeleton" interface is strongly associated with the decrease in the concentration of brain-derived neurotrophic factor (BDNF) in peripheral blood after the end of rehabilitation treatment [16]. In the above study, the decrease in the BDNF levels can be considered as a result of neurorehabilitation session. It is believed that such a decrease is indicative of active binding and internalization of this factor specifically in the nervous tissue resulting in multiple effects: axonal growth, dendrite maturation and increased synaptic plasticity.

CONCLUSIONS

The study has confirmed that children with CP show a significant ($p < 0.001$) correlation between the total ABILHAND-Kids score and the indicators of motor realization of speech production (oral praxis, pronunciation of sounds and making sentences from pictures). The complex treatment of patients with CP involving the use of the "brain–computer–hand exoskeleton" interface has resulted in the significant ($p < 0.001$) decrease in hand spasticity, the increase in the total ABILHAND-Kids score and the indicators of speech production motor realization. Such a combination of changes may be based on the plasticity processes enhancement in the neural circuits of the neocortex responsible for planning and execution of complex hand movements, as well as speech processes. The findings can be used to develop new methods for correction of motor and cognitive spheres in children with CP.

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