CASE REPORTS

Action Observation Training to Improve Activities of Daily Living and Manipulation Skills in Children with Acquired Brain Injury Secondary to an Oncologic Process: A Prospective Case Series Clinical Study

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ABSTRACT. Objective: Among solid tumours, medulloblastoma is the most common of the posterior fossa neoplasms, given that it represents 15%-20% of childhood brain tumours. The main aim of the present study was to assess the effects of action observation training on the activities of daily living (ADL) and the manipulation skills of children with acquired brain injury secondary to an oncological process. **Methods:** We recruited a consecutive convenience sample of 5 patients diagnosed with acquired brain injury secondary to an oncological process. ADL and manipulation skills were assessed using the ABILHAND-Kids and Jebsen-Taylor Hand Function Test, respectively. After conducting the initial evaluation, we planned the intervention, which lasted 10 weeks and consisted of 40 sessions for each participant, 10 in the occupational therapy department and 30 at home. After completing the intervention, we re-evaluated the main variables. **Results:** Overall, the results of the postintervention ABILHAND-Kids questionnaire showed a 5-point improvement, with a statistically significant difference and a large effect size. Eighty per cent of the sample showed better results in the total score, with differences between 4 and 8 points. In relation to manual dexterity, as measured by the Jebsen-Taylor Hand Function Test, there were no significant changes, except in one of the participants. **Conclusion:** It appears that action observation training can elicit positive changes with respect to the development of ADL, but the influence on manual dexterity was almost nonexistent.

Key words: Medulloblastoma, Action observation, Activities of daily living, Brain injury

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The Central Brain Tumor Registry of the United States reports that the incidence of medulloblastoma in patients up to 19 years of age is up to 48 for girls and 75 for boys per

100,000 patient-years and accounts for 16% of all paediatric brain tumours, with medulloblastoma accounting for 40% of all paediatric cerebellar tumours. The peak incidence of medulloblastoma occurs between 3 and 4 years of age and between 8 and 9 years of age¹⁾. Each year in Spain, 1500 new cases of cancer are diagnosed in children younger than 15 years, 15%-20% of which are central nervous system tumours. The annual incidence of these tumours is 2-5 per 100,000, with a similar distribution for both sexes²⁾.

Among solid tumours, medulloblastoma is the most

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common of the posterior fossa neoplasms, given that it represents 15%-20% of childhood brain tumours. The approach to these patients is multidisciplinary and begins with surgical resection, with the objective to establish a diagnosis and reduce the tumour volume. Subsequently, patients undergo radiotherapy and/or chemotherapy, the aim of which is to eliminate the cancer cells that persist after surgery and prevent a possible recurrence ³⁾. After the surgery phase, complications are treated in other departments, such as the paediatric intensive care unit and the rehabilitation unit. Although survival rates for this type of tumour are high, approximately 25% of survivors have motor and cognitive complications, including ataxia, balance problems, functional motor deficits, emotional lability and speech dysfunction, which typically appear between the first and second day after the surgery. Together, these symptoms comprise posterior fossa syndrome ⁴⁾.

Among the most affected functional motor deficits are manual skills and abilities and the ability to perform activities of daily living (ADL), which are addressed by occupational therapy. Although few studies have described the role of occupational therapy in treating patients with brain tumours, it is important to achieving the maximum independence possible for children in terms of their ADL. Posture, movement and practical activities are compromised in children with acquired brain damage ⁵⁾, which, together with sensory difficulties, makes it difficult for them to properly relate to their environment and they will therefore need help performing their ADL. The activities of occupational therapists are therefore essential in facilitating functional recovery, thereby helping children autonomously recover their abilities.

Movement representation techniques, such as action observation (AO) training, are a set of sensorimotor neuro-training tools widely employed in the field of neurorehabilitation ⁶⁾. AO training evokes an internal, real-time simulation of what the observer sees ⁷⁾ and can lead to an activation of areas related to the planning, adjustment and automation of voluntary movements in a manner similar to when the movement actually occurs. AO has been previously proposed for arm motor rehabilitation after stroke ⁸⁾ due to the neurophysiological properties of AO and its rehabilitative potential. Our hypothesis is that AO training can help improve ADL and manipulation skills in children with acquired brain injury secondary to an oncologic process.

The main aim of the present study was therefore to assess the effects of AO training on ADL and the manipulation skills of children with acquired brain injury secondary to an oncologic process.

Materials and Methods

Study Design

The present study was a prospective case series clinical study.

Participants

The study protocol was approved by the ethics committee of the Niño Jesús University Children's Hospital (Madrid, Spain; CI: R-0011/19). We recruited a consecutive convenience sample of 5 patients diagnosed with acquired brain injury secondary to an oncologic process, specifically a brain tumour (medulloblastoma), from the Niño Jesús University Children's Hospital. Prior to enrolment, all patients were explained the study's objectives and implications and agreed to participate by signing an informed consent document. We reviewed case records dated between October 2018 and June 2019.

The inclusion criteria for this prospective case series were as follows: age between 6 and 18 years; brain tumour diagnosis; undergoing partial or complete tumour resection surgery no more than 30 months prior to the completion of this project; undergoing oncologic therapy and standard occupational therapy at the Niño Jesús University Children's Hospital; an acquired brain injury as a consequence of the oncologic process; and a score of 1 or 2 on the Manual Ability Classification System (MACS). The exclusion criteria consisted of the following: level V in the Gross Motor Function Classification System; and the presence of moderate to severe cognitive and neuropsychological disorders according to the Wechsler Intelligence Scale for Children.

Procedures

The families attended an information session in which the project and steps to be taken in the coming months were explained. After obtaining the informed consent from the parents, we accessed each child's electronic medical records to collect the necessary information about them and their disease process. The data collected were age, gender, diagnosis, surgical interventions, neurological sequelae, use of drugs, arm involvement, dominance, cognitive level and whether they had been and/or were in a radiotherapy and/or chemotherapy programme.

The Data Protection Law (LOPD 3/2018) was always respected, as well as the European regulation (RGPD 2016/679) and rights. Once the necessary data had been collected, 2 chosen assessments were administered to determine the starting point of the patients' ADL performance, as well as the manipulation skills they presented at the start of the project.

We provided the families with the ABILHAND-Kids (AK) questionnaire. At the end of the questionnaire, the parents had to prioritise the five activities with which they felt their child had the most difficulties; these activities

Fig. 1. Intervention representation.

were addressed in the next phase of the project. If there were several activities with similar difficulty, those the parents considered most relevant in their child's environment or those of greatest importance were chosen. The Jebsen-Taylor Hand Function Test (JTHFT) was then administered.

After conducting the initial assessment, we planned the intervention, which lasted 10 weeks and consisted of 40 sessions for each participant: 10 sessions in the occupational therapy department and 30 at home, where the family conducted the same process performed by the occupational therapist in the department. Each of the 5 activities chosen by the family was performed during 2 consecutive weeks, so that the first activity was addressed during the first and second week, the second activity during the third and fourth week, the third activity during the fifth and sixth week, the fourth activity during the seventh and eighth week, and lastly the fifth activity during the ninth and tenth week. Each session lasted from 20 to 30 min. After finishing the intervention, the main variables were reassessed.

Intervention

The intervention we employed was AO training. For this purpose, a healthy boy and girl were recorded performing each of the activities in which the participants encountered difficulties. Each patient could choose whether they wanted to work with the boy's video or the girl's video during their intervention, depending on which gender they identified with more. The video was filmed from various perspectives (in third person in both the frontal and lateral plane and in first person). Figure 1 Intervention representation. shows an image of the videos employed in this preliminary study.

The video shows how the healthy boy or girl performs the complete activity and how they perform it by sequencing it in simpler motor acts. Each participant was given only those videos for the activities in which they encountered difficulties and were not given the videos of activities

that were not worked on during the intervention. Each motor act was observed for 3 min. Once all the videos of the various motor acts had been watched, the patients had to perform all the activities observed in the videos without interruption.

The AO training was combined with other Physiotherapy and Occupational Therapy interventions. These are summarised in Table 1. The Physiotherapy interventions consisted of balance and postural control training, improved motor conditioning and strength exercises, as well as the vestibular system training. Two of the patients received one Physiotherapy session per week, another patient 2 sessions, another 3 sessions and finally the last one did not receive Physiotherapy. Regarding Occupational Therapy, three of the patients did not receive any intervention and the remaining two patients received one session per week. The Occupational Therapy intervention was based on manual dexterity training and motor control tasks oriented to ADL.

Outcome Measures

ABILHAND-Kids

The AK scale assesses the perception of manual ability of children with cerebral palsy in performing the ADL determined in the scale. In total, the scale contains 21 activities classified into 4 separate levels: question mark (not applicable), impossible, difficult and easy. For the paediatric population, the questionnaire is answered by the parents, who provide their perception of their child's manual ability difficulty when performing the proposed ADL. The AK scale shows excellent internal consistency ($\alpha=0.94$) and test-retest reliability (Pearson's $r=0.91$)⁹. This scale has also shown adequate convergent validity with respect to the MACS test ($r=0.88$)¹⁰. Questionnaires that measure the functional abilities of children through their parents have been shown to be important in the evaluation of clinical changes. This is because children present a more dichotomous perception of their own abilities, allowing the measurement of ability but not of daily performance^{11,12}.

In the present study, the 21 items of the scale were not evaluated. Instead, parents were asked to indicate, among the 21 activities, the five that presented the greatest difficulties for the children, which were the therapeutic target of the subsequent intervention. Therefore, the results are shown in terms of time required to perform the activity, rather than the degree of performance. These activities could coincide among different children and, were the target of the subsequent intervention.

Jebsen-Taylor Hand Function Test

The JTHFT test measures the time it takes the child to perform each task with both the affected and unaffected arm. The objective of the JTHFT is to assess uni-manual hand functions necessary for activities of daily living. The test consists of 7 subtests, performed with both the non-

Table 1. Sample description and sociodemographic characteristics.

Case	Gender	Age	Affected upper limb	Dominance	Current chemotherapy	Pharmacological therapy	Physical therapy	Occupational therapy (out of study)	Disease staging	Grade of motor paralysis and ataxia	Presence of ocular movement disorders
1	Female	10	Right and Left	Right	Cyclophosphamide	Celecoxib Fenofibrate Thalidomide Cytarabine	1 session/week Postural control and balance treatment. Vestibular retraining	No Only AO treatment of the project	Stage IV	Hemiparesis. Severe global ataxia	Strabismus
2	Female	8	Left	Right	No	No	2 sessions/week Global and selective motor treatment. Physical conditioning, strength training	1 session/week Manual dexterity treatment, bimanual activities, task-oriented motor control for daily living tasks	Stage I	Hemiparesis. Mild global ataxia	Strabismus
3	Female	8	Left	Right	No	Valtrex	3 sessions/week Postural control and balance treatment. Vestibular retraining	1 session/week Manual dexterity treatment, bimanual activities, task-oriented motor control for daily living tasks	Stage II	Hemiparesis. Severe global ataxia	Strabismus Nistagmus
4	Male	7	Left	Right	Cyclophosphamide Etoposide	Cytarabine Neupogen Avastin Bevacizumab	No	No Only AO treatment of the project	Stage IV	Hemiparesis. Severe global ataxia	Strabismus
5	Male	11	Left	Right	No	No	1 session/week Global and selective motor treatment. Physical conditioning, strength training. Pain approach	No. Only AO treatment of the project	Stage I	Hemiparesis. No ataxia.	No

AO, action observation

dominant and dominant hand: writing, turning letters, picking up small objects and placing them in a container, stacking chips, stimulated feeding, and moving light and heavy objects¹³). In this study, all items were tested except writing. There is evidence that the JTHFT is a valid assessment tool for children aged 6 to 18 years with brain injury¹⁴). The JTHFT is reliable in a brief assessment period in children with brain injury and is responsive to changes in a brief period of intensive therapy. The minimal clinically important difference (MCID) is 54.7 s in the affected limb¹⁵).

Data Analysis

We analysed data from the 5 patients using IBM SPSS Statistics ver. 25.0 (IBM Co., Armonk, NY, USA). We employed descriptive statistics to summarise the data of the continuous variables, which are presented as mean $\pm$ standard deviation and a 95% confidence interval. We employed Student's t-test to compare the outcomes of the continuous variables, calculating the effect sizes (Cohen *d*) for the outcome variables. According to Cohen's method, the magnitude of the effect was classified as small (0.20-0.49), medium (0.50-0.79), or large (≥ 0.8). A value of $p < 0.05$ was considered statistically significant¹⁶).

Results

Our sample was composed of 5 patients (3 girls and 2 boys), with a mean age of 8.8 years (range 7-11 years) and a common medical diagnosis of medulloblastoma. After surgery, the patients presented hemiparesis, with predominant involvement of the left arm. The entire sample underwent radiotherapy and chemotherapy. Currently, none of the patients are undergoing radiotherapy, although 40% are still undergoing chemotherapy. In terms of other drugs, 60% of the sample continues with pharmacological therapy (Table 1), and none of the participants were treated with botulinum toxin. In individual terms, Table 2 shows the preintervention and postintervention data.

Case 1

For this patient, the 5 most difficult ADL were buttoning up a shirt, opening a bag of potato crisps, unwrapping a chocolate bar, opening a jam jar and unscrewing a bottle cap. After the treatment, the times for all of the activities were reduced. The most significant time differences were the activities of buttoning up a shirt and unscrewing a bottle cap, which took 56 and 98.2 s less, respectively. Moreover, after administering the AK post-treatment scale, we observed a score increase of 5 points. In terms of the JTHFT results, there was an increase in time with respect to the preintervention results, indicating no improvement in manipulation dexterity.

Case 2

The 5 ADL that the patient had the most difficulty with were buttoning up a shirt, opening a bag of potato crisps, buttoning up the trouser buttons, buttoning up a jacket and unscrewing a bottle cap. The participant managed to reduce the times for all the trained activities during the intervention, with a marked reduction in time spent buttoning up a shirt (37 s) and unscrewing a bottle cap (128.1 s). In the postintervention assessment, the increase in AK score indicated that the performance of the proposed activities had improved (by 4 points). As for the JTHFT results, there was no demonstrated improvement in manual dexterity.

Case 3

The 5 ADL that the patient had the most difficulty with were buttoning up the trouser buttons, sharpening a pencil, putting toothpaste on a toothbrush, filling a glass of water, and unscrewing a bottle cap. The time was also reduced for all activities in which the intervention was performed. There was a marked reduction in the time to button up trouser buttons (25.7 s) and unscrew a bottle cap (48.7 s). The AK results also confirmed that the performance of these activities had improved by 8 points, a result that was the highest in the entire case series. As in the previous cases, the JTHFT results showed no change in manipulation dexterity.

Case 4

The 5 most difficult ADLs for this patient were buttoning up a shirt, opening a bag of potato crisps, buttoning up the trouser buttons, sharpening a pencil and filling a glass of water. In this case, there was a considerable decrease in the time spent on 3 of the 5 activities addressed during the intervention: buttoning up the shirt, buttoning up the trouser buttons and opening a bag of potatoes (improvement of approximately 8, 6 and 1 min, respectively). In the post-intervention assessment on the AK scale, there was a 7-point increase, which confirms that the performance of the addressed activities after the intervention had improved. In terms of the JTHFT results, this is the only participant who showed reduced time spent on the test (approximately 1 min) compared with the initial assessment.

Case 5

The 5 ADLs where the patient presented the most difficulties were buttoning up a shirt, buttoning up the trousers, opening a toothpaste dispenser, sharpening a pencil, and opening a bread bag. The time spent on these tasks improved but to a lesser extent than the other cases. The activities in which the patient managed to reduce the time spent the most were buttoning up a shirt and opening a bread bag (19.5 and 10.9 s, respectively). In terms of the AK results, there were no differences compared with the re-

Table 2. Preintervention and postintervention data.

Measure	Selected AK Activities of Daily Living												AK t	JTHFT
	Button shirt	Potato bag	Trouser button	Button jacket	Unwrap chocolate	Jam jar	Bottler cap	Open tooth paste	Open bread	Remove tip	Pour tooth paste	Fill glass		
Case 1														
Initial	168.0	28.0	-	-	20.1	12.1	116.0	-	-	-	-	-	78	219.1
Middle	140.0	19.3	-	-	15.2	8.7	124.9	-	-	-	-	-		
Final	112.0	10.6	-	-	10.3	5.3	17.8	-	-	-	-	-	84	269.1
Final-Initial difference	-56.0	-17.4			-9.8	-6.8	-98.2						6	50
Case 2														
Initial	104.9	7.6	14.1	15.0	-	-	136.0	-	-	-	-	-	77	170.2
Middle	85.95	7.35	11.55	13.5	-	-	71.95	-	-	-	-	-		
Final	67.0	7.1	9.0	11.7	-	-	7.9	-	-	-	-	-	81	194.9
Final-Initial difference	-37.0	-0.5	-5.1	-3.6			-128.1						4	24.7
Case 3														
Initial	-	-	35.5	-	-	-	90.0	-	-	14.8	28.5	17.9	71	329.7
Middle	-	-	22.65	-	-	-	65.65	-	-	13.4	27.0	17.65		
Final	-	-	9.8	-	-	-	41.3	-	-	12.0	25.5	17.4	79	394.8
Final-Initial difference			-25.7				-48.7			-2.8	-3.0	0.5	8	65.1
Case 4														
Initial	694.0	82.0	359	-	-	-	-	-	-	14.4	-	26.2	65	526.7
Middle	440.5	49.45	189.5	-	-	-	-	-	-	12.5	-	25.15		
Final	187.0	19.9	20.0	-	-	-	-	-	-	9.9	-	24.1	72	464.8
Final-Initial difference	-507.0	-65.1	-339.0							-4.5		-2.1	7	-61.9
Case 5														
Initial	77.0	-	5.8	-	-	-	-	10.3	28.8	13.3	-	-	84	137.6
Middle	67.25	-	5.65	-	-	-	-	9.05	23.3	12.2	-	-		
Final	57.5	-	5.5	-	-	-	-	7.8	17.9	11.1	-	-	84	164.3
Final-Initial difference	-19.5		0.3					-2.5	-10.9	-2.2			0	26.7
Mean±SD														
Initial													75.00±7.24	276.66±157
Final													80.00±4.95	297.58±128.97
Mean difference SD (95%)													5.00±3.16	20.92±49.25
P value													0.024*	0.396

AK, ABILHAND-Kids questionnaire; AK t, ABILHAND-Kids time in seconds to perform the activity; JTHFT, Jebsen Taylor Hand Function Test; SD, standard deviation

Note: *P<0.05

sults of the preintervention assessment. As to the times obtained in the JTHFT, there was an increase compared with the pre-intervention assessment. Regarding adherence, all patients completed all OA training sessions. Adherence was complete in all cases.

Statistical Analysis

Regarding the AK variable, statistically significant pre- and post-intervention differences were found with a large effect size ($p=0.024$ $d=-0.80$; $t=2.04$) (Table 2). However, with regard the JTHFT variable, no statistically significant differences were found ($p=0.396$) (Table 2).

Discussion

The main aim of the present study was to assess the effects of AO training in ADL and manipulation skills in children with acquired brain injury secondary to an oncologic process. Overall, the results of the postintervention AK questionnaire showed an improvement of 5 points, with a statistically significant difference and large effect size. Eighty per cent of the sample showed better results in the total score, with differences between 4 and 8 points. The remaining 20% (1 child) obtained identical results for the pre-intervention and postintervention assessments. After the intervention, all the sub-tasks trained with the AO approach showed better results in terms of execution time. In relation to manual dexterity as measured by the JTHFT, there were no significant changes except in one of the participants.

All 5 participants were administered a therapeutic intervention of 20 h, distributed over 10 weeks. All participants demonstrated significant changes in trained ADL through the AO technique as measured by the AK. The study by Bleyenheuft et al. of a cohort of 80 children with unilateral cerebral palsy undergoing intensive interventions showed (through the AK questionnaire) improvements in all participants, which was greater in magnitude in the younger children (6-12 years) and with MACS levels above II⁽¹⁷⁾. Although the clinical diagnoses of the 2 studies differed, our participants had MACS levels between I and II, as well as ages between 7 and 11 years. These changes could be explained by the greater neuroplastic capacity at earlier stages of progression and at younger ages.

In addition, our results only showed significant changes in manual dexterity in 1 patient, with no correlations with ADL performance as measured by AK and manual dexterity. The study conducted by Bleyenheuft et al. showed similar results⁽¹⁷⁾. It is important to stress that the MCID for the JTHFT in children aged 13-19 years is 54.7. Given that our sample's mean age was 8.8 years (7-11 years) we cannot dispute these data. Regardless, the variations in our study participants do not reach these differences in their pretreatment and post-treatment differences, except for one of the participants, whose results exceeded

the MCID.

Another important aspect in our intervention is the focus on goals chosen by the children, increasing their motivation with task-oriented training and participation. For Novak et al., task-oriented, goal-directed training and home programs are highly recommended interventions for children with neuromotor impairment due to cerebral palsy⁽¹⁸⁾.

AO training is a sensorimotor neurotraining tool focused on provoking neurophysiological changes at the cortical level, that is, favouring a process of neuroplasticity with respect to the processes related to the planning, adjustment and automation of voluntary movements⁽⁷⁾. In this population, it is important to be able to implement these techniques, known as movement representation methods, to elicit greater neurophysiological activity at the cerebral level. A recently published umbrella review with the statistical aggregation of meta-analytic results showed that movement representation methods such as AO training and motor imagery could elicit an improvement in ADL in a population with neurological conditions⁽¹⁹⁾, which agrees with the findings in this case series study. ADL, as with any task that is amenable to a learning process, can derive benefits from AO training. For example, Cuenca-Martínez et al. found that AO training alone is an effective method for learning manual gestures for at least 4 months after completion of the intervention⁽²⁰⁾. There is ample evidence to support the use of movement representation methods to elicit a specific motor learning process⁽²¹⁻²³⁾.

The recent study conducted by Buccino et al. showed the efficacy of a 3-week intervention with AO for the rehabilitation of arm motor functions involved in ADL⁽²⁴⁾. The sample included children with cerebral palsy aged 5 to 11 years, who when treated showed increased neurophysiological activation of a parietal-premotor circuit for hand-object interactions. In addition, changes were found through neuroimaging in the left and right premotor cortex, left supramarginal gyrus and left superior temporal. Our therapeutic intervention had to be adapted to the contextual conditions and to be able to intervene in a larger number of ADL.

Limitations

This study has several important limitations that need to be considered. First, this is a case series, and all results should be taken with caution due to the small sample size. It is likely that the results could vary significantly with a larger sample. Second, due to the type of disease, the children underwent other medical interventions that might have influenced the results. We collected only the information that was reported, but this should be considered as highly important confounding variables. In addition, 1 child underwent occupational therapy outside the study, which might also influence the results, making them difficult to attribute

to the intervention alone. Finally, basic, and instrumental skills were not assessed, only manual dexterity. Future studies should assess the overall ADL for each patient.

Conclusions

Based on the results, it appears that AO training could elicit positive changes with respect to the development of ADL, but there was almost no influence on manual dexterity in the patients diagnosed with acquired brain injury secondary to an oncologic process. Although these conclusions are valid for a series of 5 cases, the results should be taken with great caution.

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