

Case Report

Multidisciplinary rehabilitation of a post-stroke pediatric patient considering the ICF perspective

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Abstract.

PURPOSE: There is a general lack of evidence on the efficacy of rehabilitation training methods after childhood stroke. The aim of the current paper is to provide an example of a multidisciplinary assessment and intensive patient-centered rehabilitation program that was devised following the Clinical Guidelines for Childhood Stroke Diagnosis, Management and Rehabilitation, based on the International Classification of Functioning, Disability and Health.

METHOD: The case of a 13-year-old teenager with physical, linguistic, cognitive and emotional impairments after acute ischemic stroke (AIS) in left middle cerebral artery territories is presented and his neurorehabilitation program is described.

RESULTS: After an intensive and comprehensive rehabilitation period, the patient showed significant improvement involving language abilities, cognitive flexibility, logical reasoning and motor independence. A 6-month post-stroke follow-up evaluation showed further gains in spontaneous language, improved motivation and collaboration, reduction of impulsiveness and better general motor stability.

CONCLUSION: This case highlights how an intensive, patient-centered, interdisciplinary rehabilitation approach can lead to good improvement across different domains, maximizing the spontaneous recovery in children and adolescents after AIS.

Keywords: Pediatric stroke, multidisciplinary rehabilitation, intensive treatment, ICF

1. Introduction

Arterial ischemic stroke (AIS) in pediatric patients is rare yet recognized as a significant cause of acquired brain injury and life-long morbidity [1]. Its incidence is progressively increasing and ranges from 1.3 to 13 per 100,000 [2]. Stroke etiology in childhood is broader than in adulthood and it is often of unknown origin [3].

Cerebral arterial dissection, which may occur spontaneously or after trauma, can be considered a rare cause of ischemic stroke in children [4].

Although ischemic stroke in children is generally perceived as having a more favorable outcome compared to adults, in a significant number of cases it leaves patients with residual impairments and long-term motor, cognitive, and behavioral consequences [5].

Recovery is a complex process occurring through a combination of spontaneous and learning-dependent processes grounded on restitution, substitution and compensation mechanisms [6]. In order to set up a specific rehabilitation program, neuropsychological and motor testing are essential to assess the presence of neu-

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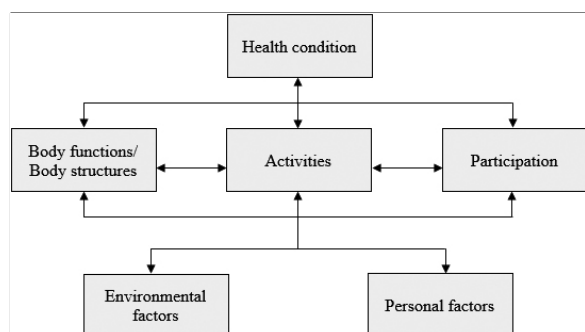


Fig. 1. Biopsychosocial model of the International Classification of Functioning, Disability and Health – Children and Youth version (ICF-CY).

22 rocognitive and/or physical impairments. Child-based
 23 rehabilitation promotes and assists neural plasticity, op-
 24 timizing recovery and outcome [7]. Taking into account
 25 all of the International Classification of Functioning,
 26 Disability and Health (ICF) domains, benefits of in-
 27 tensive and early multidisciplinary treatment are high-
 28 lighted by the Clinical Guidelines for Childhood Stroke
 29 Diagnosis, Management and Rehabilitation [8]. The use
 30 of the ICF model in clinical practice allows providers
 31 to move from purely medical or social models of dis-
 32 ability toward a biopsychosocial framework, focused
 33 on the dynamic interactions between the health condi-
 34 tion and the personal and environmental factors at play
 35 (Fig. 1) [9].

36 2. Case presentation

37 A 13-year-old Italian teenager with neither family
 38 history of neurological disease nor previous medical
 39 history except for favism disease is described.

40 On the day of the AIS, he suddenly mentioned left
 41 orbital and facial pain, fell to the ground and developed
 42 aphasia and right hemiplegia, without losing conscious-
 43 ness. There was no antecedent trauma. In the morn-
 44 ing of the onset of symptoms, headache and vomiting
 45 occurred.

46 Upon the patient's arrival to the Hospital, he had
 47 aphasia, left head and gaze deviation and right hemi-
 48 plegia. A first Magnetic Resonance Imaging (MRI) re-
 49 vealed a dissection of the left internal carotid artery
 50 and a homolateral acute ischemic territorial lesion (see
 51 Fig. 2). An attempt of mechanical thrombectomy of
 52 the occluded left Middle Cerebral Artery (MCA) was
 53 performed without success.

54 In the Intensive Care Unit, the patient was main-
 55 tained in a medically-induced coma for 5 days, while he

56 was treated with anticoagulant, antibiotic and infusion
 57 therapy. A central venous catheter and a nasogastric
 58 tube were positioned. Vital signs were within the nor-
 59 mal range, routine blood examination and ECG were
 60 normal, and EEG showed no epileptiform discharges.
 61 Five days after the onset of symptoms, another MRI
 62 confirmed a left hemisphere ischemic lesion, and a CT
 63 Angiography study showed a lack of visualization of
 64 the supraclinoid tract of the left MCA (see Fig. 3). After
 65 40 days, the anticoagulant was switched to antiplatelet
 66 therapy.

67 Tests for vasculitis, prothrombotic states, and viral
 68 neurotropic antibodies, as well as genetic analysis for
 69 Fabry disease, assessments of plasma amino acids con-
 70 centration and oropharyngeal swab were all negative.
 71 Transesophageal and transthoracic echocardiography
 72 were normal.

73 The patient was transferred to our Neurorehabilita-
 74 tion Center where he was on antiplatelet therapy. A
 75 multidisciplinary team cooperated in order to obtain a
 76 global view of his difficulties. An intensive (4–5 hours
 77 per day for 106 days) neurological rehabilitation train-
 78 ing was designed according to his specific needs and
 79 focused on speech therapy, cognitive function train-
 80 ing, motor and occupational therapy and psychological
 81 support.

82 2.1. First assessment

83 For each area of intervention, the patient underwent
 84 an initial assessment aimed at identifying a specific
 85 rehabilitation pathway.

86 A standardized clinical evaluation of his communica-
 87 tive behavior revealed the presence of motor speech
 88 disorders and severe aphasia. The patient's speech was
 89 dysarthric, almost unintelligible. Also, his spontaneous
 90 language was not fluent, characterized by word-finding
 91 impairment and by several phonemic and semantic para-
 92 phasic errors. The absence of function words and inflec-
 93 tions revealed the presence of agrammatism. Repetition
 94 and naming impairments were detected. Comprehen-
 95 sion was relatively good, with some difficulties in un-
 96 derstanding grammatical contrasts. Since the patient's
 97 motor deficits prevented him from manipulating writing
 98 instruments, the agraphia could not be assessed.

99 A comprehensive neuropsychological examination
 100 highlighted the presence of several difficulties in
 101 problem solving, planning, cognitive flexibility, self-
 102 monitoring and cognitive inhibition. Mild deficits were
 103 found in the nonverbal intelligence domain. No im-
 104 pairments of perception, memory or attention were ob-
 105 served.

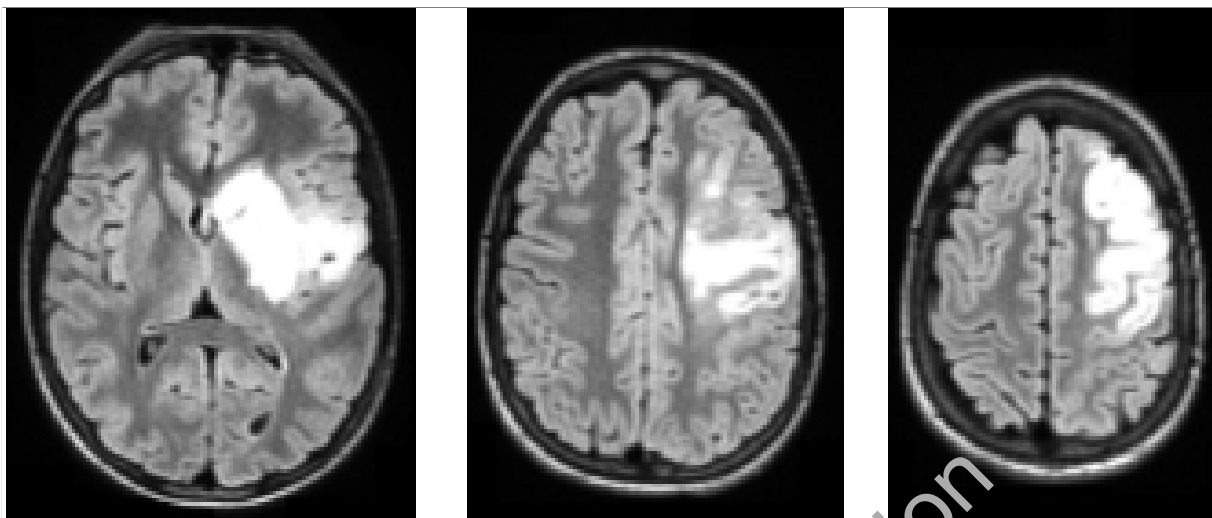


Fig. 2. Brain magnetic resonance imaging showing the acute left ischemic lesion on FLAIR axial scans.



Fig. 3. Brain CT angiography revealing left internal carotid artery occlusion.

The patient presented with severe right hemiplegia with right upper limb hypotonia. He was only partially autonomous in postural changes. Thus, he was completely dependent in basic (BADL) and instrumental (IADL) activities of daily living, needing physical assistance. Motor tests showed a good performance in static sitting balance and more difficulties in dynamic sitting balance and coordination.

Specific linguistic, cognitive and motor tests with raw scores and percentages of accuracy are reported in Table 1.

With respect to behavioral and psychological response, the patient struggled to establish a good working alliance with all of the specialists, showing avoidance signals and emotional unavailability. He presented

consistent anxiety-related behaviors, psychological distress and poor motivation, and refused complex exercises in order to avoid frustrating experiences. In addition, he showed poor self-reflection ability, attempting to escape from negative thoughts and feelings.

2.2. Intervention

After the initial speech evaluation, a twice daily rehabilitation program was focused on articulation, verbal production and auditory and reading comprehension. For specific tasks, see Table 2.

Compensatory approaches for aphasia intervention in adult patients, such as the Promoting Aphasics' Communicative Effectiveness (PACE) [19] and the application of Semantic Feature Analysis (SFA) [20], were adapted to the patient's needs. Moreover, a smartphone and tablet were used to facilitate communication.

A specific daily neuropsychological rehabilitation program was designed with paper and pencil exercises and computerized tasks using the rehabilitation software "ERICA" [21]. Cognitive training was focused mainly on executive functions, while sustained, selective and divided attention tasks were aimed at reducing impulsivity (see Table 2). The complexity of each exercise was progressively increased to match the patient's improvement in accuracy and processing speed and decrease of impulsiveness.

The motor impairment was treated with physiotherapy sessions twice daily. Exercises were aimed at strengthening muscles, decreasing spasticity, improving transferring abilities and postural changes, increas-

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Table 1
Linguistic, cognitive and motor scores at admission, discharge and follow-up

Test	Max	1 st assessment Raw score (% of accuracy)	2 nd assessment Raw score (% of accuracy)	3 rd assessment Raw score (% of accuracy)
Language				
<i>AAT</i>				
Token test, errors	150	20 (13%)	5 (3%)	5 (3%)
Repetition	150	112 (75%)	120 (80%)	126 (84%)
Written language	90	na	na	77 (86%)
Naming	120	76 (63%)	109 (91%)	110 (92%)
Comprehension	120	108 (90%)	110 (92%)	113 (94%)
<i>TROG-2</i>				
Block passed	20	10 (50%)	14 (70%)	14 (70%)
Executive functions				
<i>WCST</i>				
Number of categories achieved	6	4 (67%)	6 (100%)	6 (100%)
Perseverative responses	128	18 (14%)	8 (6%)	13 (10%)
Non-perseverative errors	128	39 (30%)	36 (28%)	5 (5%)
<i>ToL</i>				
Correct responses	36	31 (86%)	32 (89%)	29 (80%)
Initiation time (in seconds)	–	10	97	55
Execution time (in seconds)	–	151	113	117
Total time (in seconds)	–	161	212	172
Nonverbal logic reasoning				
SPM		31 (52%)	41 (68%)	41 (68%)
Apraxia				
<i>Italian orofacial apraxia test</i>				
Total on verbal request	36	18 (50%)	27 (75%)	30 (83%)
Total on imitation	36	26 (72%)	36 (100%)	36 (100%)
Dysarthria				
<i>Robertson dysarthria profile</i>				
Respiration	20	12 (60%)	15 (75%)	17 (85%)
Phonation	48	39 (81%)	41 (85%)	47 (98%)
Facial musculature	80	55 (69%)	67 (84%)	80 (100%)
Diadochokinesis	44	0 (0%)	25 (57%)	20 (45%)
Reflexes	28	26 (93%)	27 (96%)	28 (100%)
Articulation	20	7 (35%)	12 (60%)	14 (70%)
Intelligibility	24	7 (29%)	13 (54%)	15 (62%)
Prosody	20	7 (35%)	13 (65%)	13 (65%)
TOT	284	153 (54%)	213 (75%)	234 (82%)
Motor function				
TIS	23	15 (65%)	16 (70%)	17 (74%)
FAC	6	2 (30%)	5 (83%)	5 (83%)

na = non administrable. AAT = Aachen Aphasia Test [10]; TROG-2 = Test for Reception of Grammar 2 [11]; WCST = Wisconsin Card Sorting Test [12]; ToL = Tower of London [13]; SPM = Standard Progressive Matrices [14]; Italian Orofacial Apraxia Test [15]; Robertson Dysarthria Profile [16]; TIS = Trunk Impairment Scale [17]; FAC = Functional Ambulatory Classification [18].

ing balance and posture control during standing and walking, walking indoors and outdoors independently and climbing up and down stairs. See Table 2 for specific tasks. A carbon fiber ankle-foot orthosis (AFO) and a nighttime brace for the right upper limb were adopted to maintain a full active range of motion and prevent contractures. In addition to traditional physical therapy, electrostimulation, virtual reality activities and hydrokinetic therapy were performed.

Two occupational therapy sessions per day were aimed at improving fine upper limb motor skills in order to enhance the patient's participation and increase his independence during ADLs.

Psychological support was provided in twenty sessions and focused on cognitive processing of the patient's traumatic experience. Imaginative techniques and metacognitive strategies were taught to decrease restlessness and support him during his difficult rehabilitation training. The last sessions were dedicated to the return of the patient into society and school. During the sessions, he established a good therapeutic alliance, with increasing self-disclosure and emotional openness over time. His use of defense mechanisms was gradually reduced, which allowed for greater availability and cooperation in working with all of the team's specialists.

Table 2 Linguistic (A), cognitive (B), physical (C) and occupational (D) therapy specific tasks			
Speech and language therapy (A)	Cognition therapy(B)	Physical therapy (C)	Occupational therapy (D)
<p><i>Articulation:</i> reading and repetition of isolated sounds, words of increasing length and complexity and sentences with increasing numbers of constituents, active and passive praxis exercises, oral-motor exercises with and without using mirror, strategies to improve the speech intelligibility (hyper-articulation and slowing speech rate)</p> <p><i>Verbal production:</i> pictures naming, description tasks, semantic jokes and riddles, semantic and phonological fluency exercises, storytelling</p> <p><i>Auditory and reading comprehension:</i> comprehension of spoken and written words and sentences of increasing length and complexity, reading comprehension exercises with multiple choices questions, yes/no questions, scrambled-sentence tasks</p>	<p><i>Executive functions:</i> verbal and visuospatial problem-solving tasks (decomposing tasks into sub-goals, generating alternative solutions to problems, shifting between ideas and actions, monitoring ongoing performances), rule-deduction tasks, shifting games and training for the inhibition of automatic behaviors and impulsiveness</p> <p><i>Sustained, selective and divided attention:</i> dual-task exercises, attention demanding tasks with moving targets, Go/No-Go tasks with increasing number of distractors</p>	<p><i>Lower limb:</i> repetitive movements (hip flexion and external/internal rotation, knee extension, ankle flexion), muscle contractions, flexibility exercises, muscles tone management, supported standing balance activities, walking on level and unlevel surfaces, avoiding obstacles, climbing up and down stairs</p> <p><i>Upper limb:</i> arm, wrist and hand stretch, reaching, grasping, pitching, gripping and releasing activities, active and passive mobilization exercises, coordination and bi-manual tasks</p>	<p><i>Autonomies:</i> training to improve occupational performance in BADL (feeding, grooming, bathing, upper and lower body dressing) and IADL (using telephone, tablet handwriting) in different contexts of daily life (home, school), counseling on environmental adaptations and daily life aids to facilitate occupational performance and enhance independence</p> <p><i>Fine upper limb motor skills:</i> ecological exercises aimed at reaching, gripping, manipulating small objects, bi-manual activities, hand and finger coordination tasks, specific exercises to reduce spasticity and enhance muscles strength</p> <p><i>Participation:</i> games, daily living activities and role playing to enhance awareness and promote active participation</p>
177	2.3. <i>Final assessment</i>	178	203
178	At discharge, the patient showed general improve-	179	204
179	ment. Advancement was particularly evident in verbal	180	205
180	comprehension and understanding of grammatical con-	181	206
181	trasts, as well as in naming and articulation abilities.	182	207
182	Due to better speech intelligibility, a significant gain in	183	208
183	spontaneous language was observed, with an increased	184	
184	communicative intent that included conversations on	185	
185	topics of everyday life.	186	
186	A second neuropsychological assessment showed	187	
187	better nonverbal logical reasoning and an increased	188	
188	ability to coordinate thoughts and behavior, with more	189	
189	cognitive flexibility, as well as improved planning and	190	
190	self-monitoring skills.	191	
191	Regarding motor skills, the patient reached good au-	192	
192	tonomy in postural changes and walking ability, requir-	193	
193	ing supervision only on uneven surfaces. If helped by	194	
194	supervisors with verbal reminders, he was more inde-	195	
195	pendent in BADL and IADL.	196	
196	Results of the final assessment are summarized in	197	
197	Table 1.	198	
198	With respect to the emotional domain, the patient	199	
199	demonstrated greater reduction of impulsiveness, good	200	
200	availability and cooperation with all of the team's spe-	201	
201	cialists, increased emotional availability and reduction	202	
202	of social withdrawal.		
		After discharge, the patient was reintegrated into	203
		home and school life with environmental support (e.g.	204
		orthotic devices for mobility, optimal arrangement of	205
		furniture, digital tablet for communication, daily living	206
		aids). He attended the last months of middle school and	207
		passed his eighth grade finals.	208
		2.4. <i>Follow-up assessment</i>	209
		A 6-month post-stroke follow-up assessment was	210
		performed after a period of sub-intensive (three days	211
		a week) speech therapy and neuro-motor training in	212
		a different structure. The evaluation showed a general	213
		stability in both linguistic and motor domains, with fur-	214
		ther mild improvement in all tests. The executive func-	215
		tions, in particular planning and self-monitoring abil-	216
		ities, were the only areas in which the patient showed	217
		a mild decrease in performance compared to his dis-	218
		charge evaluation. Written language assessment high-	219
		lighted the presence of mild agraphia. For raw scores	220
		and percentages of accuracy, see Table 1.	221
		3. Discussion	222
		The aim of this paper is to report the multidisciplinary	223
		assessment and the patient-centered rehabilitation pro-	224

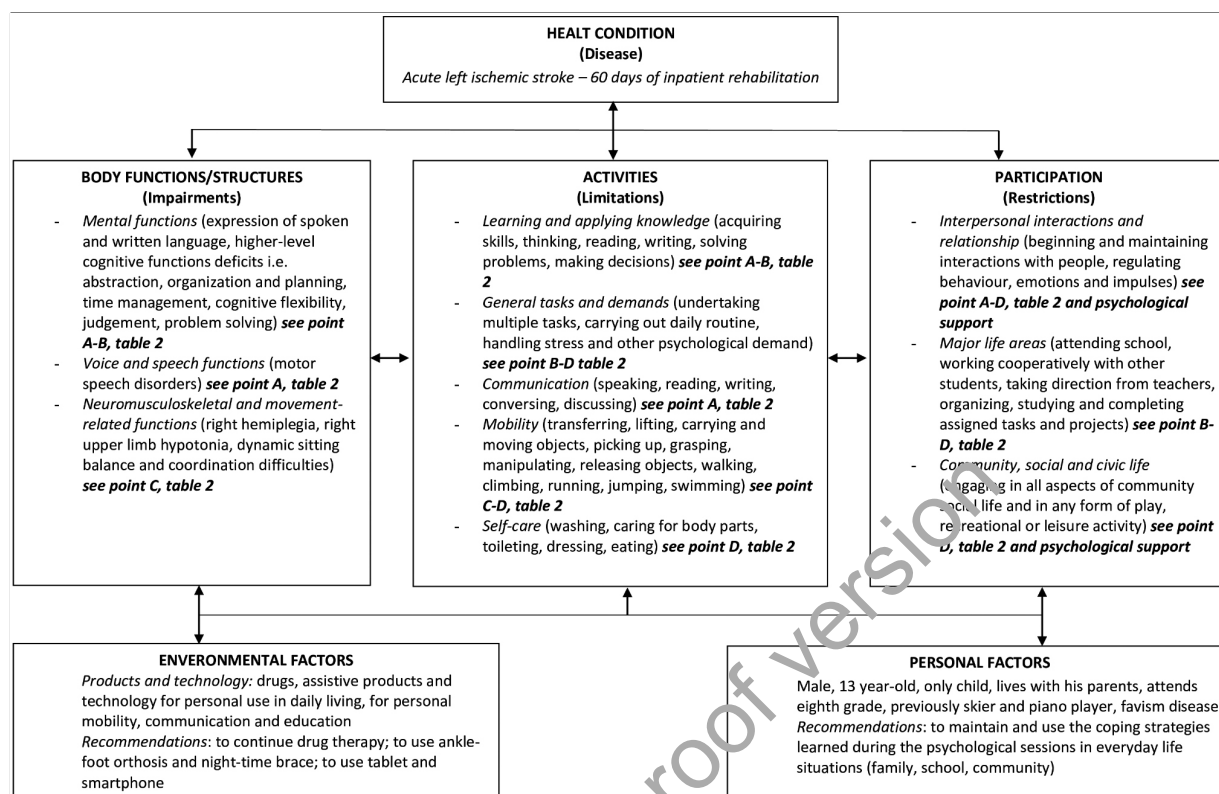


Fig. 4. Modified ICF-CY model considering the patient's characteristics and his rehabilitation program.

gram of a 13-year-old AIS patient who subsequently made significant clinical improvement. As the Clinical Guidelines for Childhood Stroke Diagnosis, Management and Rehabilitation suggest, an early, intensive and multimodal intervention was developed [8]. Referring to the ICF model, the patient functioning was considered in terms of the dynamic interactions between his disability and environmental and personal factors (Fig. 4). The ICF, supporting both medical and social models of human functioning and disability, provides a multidimensional framework that can be used to set up comprehensive and interdisciplinary approaches to assessment and intervention (Tables 1 and 2 and Fig. 4). Moreover, it offers a common language for health, social and educational services that can be used across different settings and disciplines [9].

Because descriptions of post-stroke rehabilitation training in childhood are rare, we referred to adult-stroke literature, adapting the therapeutic environment to the patient's specific needs in terms of modality, intensity and location of the intervention.

Since children and adolescents usually show consistent difficulties working in structured settings, which are required in adult standard training [22], it is im-

portant to customize the treatment in accordance to the young patient's needs [23]. All therapeutic interventions were tailored to the teenager's specific profile. Therapists tried to develop a good working alliance with him by including some enjoyable tasks in each session. Literature highlights how high levels of patient engagement seem to be strongly associated with better rehabilitation outcomes [6]. In agreement with these results, consistent improvements in all rehabilitation tasks were observed as the patient became more interested, cooperative and involved in training.

Speech therapy sessions followed scientific recommendations for adult aphasic patients [24] and were adapted to the teenager's characteristics. As literature suggests, familiar technology was incorporated for a more functional treatment [22]. After the rehabilitation period, the patient's language improved, even more clearly at the follow-up evaluation, where he showed greater communicative intent and a general reduction of inhibition.

Concerning cognition, substantial evidence supports the effectiveness of cognitive rehabilitation after acquired brain lesions, highlighting how neuropsychological treatment can help patients learn and compensate

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for residual limitations in this area [24]. Cognitive tasks were mainly assigned to the patient using the software ERICA [21] and included computerized exercises based on advanced cognitive rehabilitation models. Problem-solving tasks were administered to improve executive functions while attention exercises were aimed at reducing impulsivity. The presence of executive functioning deficits is known to moderate and negatively affect the overall response to treatment [24]. Moreover, during adolescence, the brain is still changing and the frontal cortex, classically associated with the control of executive functions, is partially immature and still developing in an injured brain. Cognitive tasks require patients to create new strategies and consequently new neural circuits that replace damaged ones. During treatment, the complexity of each exercise was progressively increased as the patient showed better cognitive flexibility and self-monitoring skills. The patient's profile showed a partial reappearance of difficulties after a period without any training for cognitive functions. This is consistent with other case descriptions that indicate nonsignificant decrements of performance between discharge and follow-up assessments [25]. Hence, we can infer that intensive treatment is effective and should be long-term in order to maintain results over time.

Physical and occupational therapies were individualized and exercises followed a task-oriented approach. Also, traditional physical therapy was utilized. Electrostimulation and hydrokinetic therapy were introduced too. Following recent findings [26], virtual reality activities were added and were well accepted by the teenager. At discharge, the patient showed general motor improvements, reaching good autonomy in postural changes, gait and ADLs. His advances remained largely unchanged at follow-up. Increased emotional availability and cooperation were observed over time with all of the team's specialists.

In conclusion, this case highlights how an early, intensive and comprehensive multidisciplinary rehabilitation program following the Clinical Guidelines for Childhood Stroke Diagnosis, Management and Rehabilitation can lead to significant improvement in children after AIS. In accordance with the ICF model, the treatment was tailored to the patient's specific needs, environment and personal factors, in order to obtain the best outcome possible and allow the child and his family to achieve a higher quality of life. Further studies with a larger number of patients are needed to support evidence of efficacy of childhood stroke rehabilitation related to Clinical Guidelines.

Acknowledgments

The authors would like to thank the patient and his family, as well as the World Health Organization for the permission to reproduce their material. All authors gave their consent to paper submission and approved the current version of the paper. They agreed to take public responsibility for all the paper's content. The first two authors contributed equally to the work. In addition, the authors ensured that they have written entirely original work and appropriately cited or quoted the considered studies. No financial support for the research, authorship and/or publication of this article was received.

Conflict of interest

The authors have no conflict of interest to report.

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