



Early rehabilitation improves neurofunctional outcome after surgery in children with spinal tumors

Nezire Kose¹, Ozge Muezzinoglu¹, Sevil Bilgin¹, Sevilay Karahan², Ilkay Isikay³, Burcak Bilginer³

¹ Department of Physiotherapy and Rehabilitation, Faculty of Health Sciences, Hacettepe University, Ankara, Turkey

² Department of Biostatistics, Faculty of Health Sciences, Hacettepe University, Ankara, Turkey

³ Department of Neurosurgery, Faculty of Medicine, Hacettepe University, Ankara, Turkey

Corresponding author:

Nezire Kose, P.T., Ph.D., Department of Physiotherapy and Rehabilitation, Faculty of Health Sciences, Hacettepe University, Ankara, Turkey, fztnezire@yahoo.com.
Sevil Bilgin, P.T., Ph.D., Department of Physiotherapy and Rehabilitation, Faculty of Health Sciences, Hacettepe University, Ankara, Turkey, sevilcuvalci@yahoo.com.

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Abstract

To investigate the effect of early rehabilitation on neurofunctional outcome after surgery in children with spinal tumors, this study reviewed the medical charts and radiographic records of 70 pediatric patients (1–17 years old) who received spinal tumor surgical removal. The pediatric patients received rehabilitation treatment at 4 (range, 2–7) days after surgery for 10 (range, 7–23) days. Results from the Modified McCormick Scale, Functional Independence Measure for Children, American Spinal Injury Association Impairment Scale and Karnofsky Performance Status Scale demonstrated that the sensory function, motor function and activity of daily living of pediatric children who received early rehabilitation were significantly improved. Results also showed that tumor setting and level localization as well as patients's clinical symptoms have no influences on neurofunctional outcomes.

Key Words: nerve regeneration; spinal cord injury; spinal cord; tumor; children; rehabilitation; function; sense; motor; retrospective analysis; neural regeneration

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Introduction

In children, primary spinal tumors comprise 4–8% of all tumors from the central nervous system. They are rarely found in pediatric patients, with a reported incidence rate of 1–2.6 per one million children around the world^[1].

The gross classification of tumors based on anatomic location can be divided into three discrete areas. First, the extradural tumors localize to the area between the bony structures and the dura mater. Second, the intradural tumors are subdivided into extramedullary and intramedullary. Intradural extramedullary tumors occur in the area within the dura but not part of the spinal cord. Intradural intramedullary tumors are seen within the spinal cord parenchyma^[2-3].

Spinal tumors can present with a variety of clinical signs and symptoms in children such as pain followed by motor regression, gait disturbance, sphincter dysfunction, sensory symptoms, torticollis, and kyphoscoliosis^[4-7].

Treatment of spinal tumors is based on tumor type, but surgical resection is the mainstay^[2]. Conventionally, conservative management consists of biopsy, dural decompression, chemotherapy, and radiation therapy for spinal tumors^[7-8]. Moreover, physical treatment and rehabilitation implementation are necessary in order to minimize the symptoms of the patients and develop present functions. The aims of rehabilitation programs are to decrease the severity of the

symptoms, raise the levels of functional independency and life quality, and prevent possible complications. Rehabilitation programs should include the provision of mobility (ambulation, use of wheelchair, transport methods) and daily life activities (dressing, hygiene and self-care activities), sphincter control training, prevention and treatment of decubitus ulcers, treatment of pain and spasticity, postural training, and determination of and training for necessary equipment for mobilization and home programs^[9-12].

Although there have been many studies about primary treatment (surgery, radiation therapy, chemotherapy) results in adult patients with spinal tumors^[11-20], there are a few studies about early rehabilitation in children following spinal tumor surgery^[11, 21]. The purpose of the current study was to investigate the effect of early rehabilitation on neurofunctional outcome after surgery in children with spinal tumors.

Results

Patient characteristics

Seventy pediatric children were included in this study. Their general characteristics are shown in Table 1. The most common presenting symptoms were limb weakness (100%). The types and locations of spinal tumors and tumor setting are shown in Table 2. Of these patients, 49% had intradural, extramedullary spinal tumors.

Table 1 Patient characteristics (n = 70)

Age (years)	
Mean ± SD	10.9±5.4
Minimum–maximum	1–17
Sex [n(%)]	
Male	39(55)
Female	31(45)
Tumor setting [n(%)]	
ED	20(29)
IDEM	16(22)
IDIM	34(49)
Clinical symptoms prior to rehabilitation [n(%)]	
Pain	39(56)
Urinary incontinence	17(24)
Weakness or paralysis of the extremities	70(100)
Monoparesis	3(4)
Paraparesis	42(60)
Quadriparesis	13(19)
Paraplegia	8(11)
Hemiparesis	4(6)
Sensory deficit	
Anesthesia	3(4)
Not evaluated	11(16)
Hypoesthesia	40(57)
Normal	16(23)
Length of stay in neurosurgery unit (day)	
Median (IQR)	18(13–34)
The onset-to-admission interval (day)	
Median (IQR)	4(2–7)
Duration of rehabilitation (day)	
Median (IQR)	10(7–23)

SD: Standard deviation; ED: extradural; IDEM: intradural extramedullary; IDIM: intradural intramedullary; IQR: interquartile range.

Neurofunctional outcomes

Neurological assessments were based on American Spinal Injury Association Impairment Scale (AIS) score-determined spinal cord function. Pre- and post-rehabilitation AIS scores and the change of scores are presented in Table 3. One patient who presented with AIS score D impairment improved to score E function, and 24 remained with neurological deficits (AIS score D). Nine patients who presented with AIS score C impairment improved to score D function and that development was statistically significant ($P < 0.05$). One patient with score B impairment improved to score C, while six patients with AIS score A impairment remained with neurological deficits.

The changes in pre- and post-rehabilitation Modified McCormick Scale (MMS), Functional Independence Measure for Children (WeeFIM), and Karnofsky Performance Status Scale (KPS) scores were statistically significant ($P < 0.05$) (Table 4). The percentage changes of MMS and WeeFIM scores were not significantly correlated with the percentage changes of KPS scores and onset-to-admission interval (the time between surgery and admission to rehabilitation). However, there was a negative correlation between the percentage change of MMS score and age, and a positive correlation between percentage change of KPS scores and age ($r = -0.30$, r

$= 0.31$, $P < 0.05$). There was no correlation between the percentage change of WeeFIM score and age ($r = 0.19$, $P > 0.05$). These results indicated that neurofunctional recovery was better in older children. Length of stay in neurosurgery unit (LOS) was positively correlated with the percentage change of WeeFIM and KPS score ($r = 0.54$, $r = 0.03$, $P < 0.05$). The duration of hospitalization was positively correlated with the percentage change of WeeFIM and KPS scores ($r = 0.62$, $r = 0.32$, $P < 0.05$). There was no significant correlation between the percentage changes of MMS and LOS and the duration of hospitalization ($r = -0.11$, $r = -0.22$, $P > 0.05$) (Table 5). Sex, tumor setting and level of localization, recurrence, radiotherapy, chemotherapy, orthopedic problems, clinical symptoms pre-rehabilitation were not correlated with the percentage change of neurofunctional outcomes ($P > 0.05$).

Discussion

The main sign and symptoms that characterize spinal cord compression are pain, weakness, autonomic dysfunction, and sensory loss. Clinical complications of this condition include paralysis of muscles innervated caudal to level of spinal cord involvement, impairment of bladder, bowel, and sexual function, pain, and a decrease in functional mobility and self-care skills^[11]. In our study, there was limb weakness in 100%, sensory deficits in 71%, localized or dermatomal pain in 56% and urinary incontinence in 24% of pediatric patients.

There were a few studies in the literature with regard to children with spinal tumors who underwent rehabilitation in the early stages following surgery. Rehabilitation is an essential component in the management of pediatric spine and spinal tumors^[10, 21]. This should begin as soon as the child is clinically well enough and any potential spinal instability has been managed (surgical fixation/bracing). Regular physiotherapy should be used to maximize mobility, and early occupational therapy is essential both to provide equipment and to undertake an assessment of the child's home if their mobility has been significantly impaired^[10]. Mora and Woollner^[21] indicated that when the epidural non-Hodgkin lymphoma is treated aggressively with systemic chemotherapy and vigorous rehabilitation, the prognosis for complete neurological recovery and cure of the lymphoma are also encouraging.

Dincer and colleagues^[22] included 98 children who were operated on for spinal tumors and had neurological problems in their study. The patients were administered an intense physical treatment program including passive, active, resistance, and tension exercises, and ambulation and sphincter control training. They monitored the patients for 1–15 months and reported independence in daily life activities and ambulation in the group with severe neurological problems. In the group with very severe neurological problems, some patients could walk with suitable orthosis, but some of them still needed wheelchairs. Choksi et al.^[23] demonstrated neurofunctional recovery in mobility and self-care from admission to discharge during inpatient rehabilitation at the Franciscan Hospital for Children in 32 children with spinal cord injury between 1 and 19 years of

Table 2 Setting and histologic types of spinal tumors (n = 70)

Histology	n	Level of localization					Tumor setting			
		C	CT	T	TL	L	LS	IDIM	IDEM	ED
Aneurysmal bone cyst	2			2						2
Astrocytoma	17	2	7	5	3			16	1	
Dermoid cyst	1		1					1		
Epandymoma	12	2		4	6			9	3	
Epidermoid cyst	4		1	1	1	1		3	1	
Ewing sarcoma	2			1				1		2
Ganglioneuroma	3		1			2				3
Germ cell tumor	1			1						1
Hemangioma	4			4						4
Histiocytosis X	1						1		1	
Lipoma	2		1	1				2		
Non-Hodgkin's lymphoma	3			3						3
Neuroblastoma	6	1		2			2		1	4
Neurofibroma	3	2		1					3	
Schwannoma	7	3	1	2		1			6	1
Cervical congenital cyst	1		1					1		
Teratoma	1		1					1		
Wilm's tumor	1			1						1
Total [n(%)]		10(14)	14(20)	28(40)	10(14)	4(6)	4(6)	34(49)	16(23)	20 (28)

C: Cervical; CT: cervicothoracic; T: thoracic; TL: thoracolumbal; L: lumbal; LS: lumbosacral; IDIM: intradural intramedullary; IDEM: intradural extramedullary; ED: extradural.

Table 3 Pre- and post-rehabilitation neurological status according to the American Spinal Injury Association Impairment Scale classification

		Post-rehabilitation					Total
		A	B	C	D	E	
Pre-rehabilitation	A	6	0	0	0	0	6
	B	0	2	1	0	0	3
	C	0	0	27	9 ^a	0	36
	D	0	0	0	24	1	25
	E	0	0	0	0	0	0
Total		6	2	28	33	1	70

Superscript "a": Marginal homogeneity test, $z = -3.317$, $P = 0.001$.

age. Children typically received therapy for a minimum of 3 hours per day, including individual physical therapy and occupational therapy. The children showed both improvement in neurofunctional skills and reduction in caregiver assistance across mobility and self-care domains. These findings were consistent with previous studies by Garcia et al.^[24] and Allen et al.^[25], which reported significant improvement in functional status of children with spinal cord injury after inpatient rehabilitation. In our study, we also found that the neurofunctional outcomes of patients significantly increased.

Neurological recovery is an important aspect of rehabilitation in patients with spinal tumors^[26]. According to the AIS impairment scale, significant neurological recovery was noted in this study after early rehabilitation. Eleven patients showed recovery by at least one grade by the time of discharge. The results were similar to the study of Gupta et al.^[27], who reported 106 patients with traumatic or nontraumatic spinal cord injury administered rehabilitation at least 2 hours daily for 14–193 days. They achieved at least one

grade of improvement in AIS scores in 51% of their rehabilitation group.

We investigated the correlations between age, tumor setting and level localization, clinical symptoms pre-rehabilitation, the duration of hospitalization/rehabilitation, the onset-to-admission interval and functional outcomes. We demonstrated a significant correlation between the percentage changes of WeeFIM and KPS scores and the duration of rehabilitation. Functional recovery was faster if the rehabilitation was longer. Rehabilitation strategies should target cortical plasticity and promote successful representational competition of neurons with direct and indirect inputs to the affected spinal motor pools from the remaining cortex. It is known that task-specific treatment often leads to better performance and cortical representational plasticity by a growth-related mechanism, such as increasing numbers of dendritic spines, axonal and dendritic branching, and synaptogenesis^[28].

We did not find any significant correlation between the

Table 4 MMS, KPS and WeeFIM scores prior to and after rehabilitation

	Pre-rehabilitation		Post-rehabilitation		P ^a
	mean±SD	Minimum-maximum	mean±SD	Minimum-maximum	
MMS	3.6±0.8	2–5	3.4±1.0	1–5	< 0.001
KPS	51.6±10.9	3–80	54.9±14.1	10–90	< 0.001
WeeFIM	43.8±21.0	13–90	49.0±20.8	13–91	0.002

MMS: Modified McCormick Scale; KPS: Karnofsky Performance Status Scale; WeeFIM: The Functional Independence Measure for Children; SD: Standard deviation. Superscript "a": Wilcoxon signed-rank test, $P < 0.05$.

Table 5 Correlation of age, LOS and duration of rehabilitation with OAI and the percentage change of MMS, WeeFIM, and Karnofsky scores

	MMS change		WeeFIM change		KPS change	
	r	P	r	P	r	P
Age	-0.30	0.01 ^a	0.19	0.12	0.31	0.01 ^a
LOS	-0.11	0.38	0.54	< 0.001 ^a	0.26	0.03 ^a
Duration of rehabilitation	-0.22	0.06	0.62	< 0.001 ^a	0.32	0.01 ^a
OAI	0.14	0.24	0.03	0.83	-0.14	0.24

MMS: Modified McCormick Scale; WeeFIM: The Functional Independence Measure for Children; KPS: Karnofsky Performance Status Scale; LOS: length of stay in neurosurgery unit; OAI: the onset-to-admission interval; r: correlation coefficient (Spearman correlation coefficient) ^a $P < 0.05$.

percentage changes of MMS, WeeFIM and Karnofsky scores and the onset to admission interval. We suppose that the reason was the relatively short duration of onset to admission interval (2–7 days) and that neurofunctional recovery might be later in children with longer onset to admission intervals. In addition, we found a negative correlation between age and the percentage change of MMS and a positive correlation between age and the percentage change of KPS score. We suggested that neurofunctional recovery was faster in older children who participated in rehabilitation programs better. Syczewska et al.^[29] reported that the age of illness onset influenced the severity of functional status. The median value and the upper limit of gait parameters for the spinal tumor patients were the lowest values, suggesting that this type of tumor appeared at an earlier age. Despite this, there was no significant relationship between age and the percentage change of WeeFIM score. This finding was considered to be due to postoperative fear sensation in children and inability to show their independence as a result of the protective attitude of their parents.

At the same time, we evaluated whether there was a difference between other risk factor groups (sex, tumor setting and level localization, recurrence, radiotherapy, chemotherapy, orthopedic problems, clinical symptoms pre-rehabilitation) according to the percentage change of neurofunctional outcomes (WeeFIM, MMS, and KPS scores). We did not find any correlation between these parameters. Thus, we concluded that rehabilitation programs should be initiated as soon as possible without considering tumor setting and level localization or clinical symptoms.

Rehabilitation has an indispensable role to play in favorable neurofunctional outcome and prevention/management of symptoms after spinal tumors. Rehabilitation programs initiated earlier are effective in achieving functional independence levels and recovery of neurological functions. In these

patients, rehabilitation programs should be administered for longer periods, suitable home exercise programs should be prepared, and the contribution of family members in early periods should be enabled. The limited number of studies that investigate rehabilitation outcomes in children with spinal tumors should be increased. We think that our study will lead the way for further studies.

Subjects and Methods

Subjects

This study retrospectively reviewed the medical charts and radiographic records of 70 pediatric patients with vertebral or spinal tumors who received surgical treatment in Department of Neurosurgery, Hacettepe University Hospital, Turkey between 1992 and 2012. All patients were 18 years or younger at the time of admission.

Methods

Patient characteristics

Baseline epidemiological characteristics (sex, age), tumor-related data (tumor setting and level localization, histology), clinical symptoms pre-rehabilitation, length of stay in neurosurgery unit, onset-to-admission interval (the time between surgery and admission to rehabilitation), and duration of rehabilitation were retrospectively analyzed.

Neurofunctional assessments

Patients were evaluated with the MMS, WeeFIM, AIS, and KPS scales before and after rehabilitation. The MMS, a method of assessing tumor impact on sensory and motor functions, is a tool frequently found in the neurosurgical literature for evaluating patients with intramedullary tumors. This functional outcome tool has been modified for use in evaluating children with tumors (Table 6)^[2].

Table 6 Modified McCormick Scale for functional classification in pediatric patients

Grade	Definition
1	Neurologically intact, ambulate normally, may have minimal dysesthesia
2	Mild motor and sensory deficit, maintain functional independence
3	Moderate deficit, limitation in function, independent with external aid
4	Severe motor or sensory deficit, depend on external assistance
5	Paraplegia or quadriplegia

The WeeFIM is an instrument used to determine the degree of disability in basic daily living. It comprises 18 items grouped into two subscales: the motor subscale including self-care (6 items), sphincter control (2 items), transfers (3 items), and locomotion (2 items); and the cognitive subscale including communication (2 items) and social cognition (3 items). A 7-level ordinal rating system ranging from 7 (complete independence) to 1 (total assistance) was used to rate the performances^[30-31]. As none of our patients had cognitive impairment, we used only the motor subscale of the WeeFIM in our study.

The AIS impairment scale runs from A to E, at the time of injury and 3 months afterwards: AIS-A (no motor or sensory function is preserved in the sacral segments S₄₋₅), AIS-B (sensory but not motor function is preserved below the neurological level and includes the sacral segments S₄₋₅), AIS-C (motor function is preserved below the neurological level and more than half of the key muscles below the neurological level have a muscle grade less than 3), AIS-D (motor function is preserved below the neurological level and more than half of the key muscles below the neurological level have a muscle grade of 3 or above) and AIS-E (motor and sensory functions are normal). Grade A or B of AIS indicates that the patients have complete motor palsy^[32].

The KPS Scale is the most widely used method of quantifying the functional status of cancer. It is an 11-point rating scale that ranges from normal functioning (100) to dead (0)^[33].

Physical therapy and rehabilitation program

The patients were included in the rehabilitation program as soon as their medical conditions were stabilized following surgery in the Neurosurgery Department. The median of the onset to admission interval was 4 (2–7) days and the median of duration of rehabilitation was 10 (7–23) days. These patients underwent programs that were conducted for pulmonary rehabilitation, improvement in or complete recovery from pain and spasms, regulation of muscular tonus, prevention of the complications of decubitus positions and improvement or treatment of existing ones, progressive neuromuscular facilitation, sensation-perception training, and training for daily life activities in accordance with their medical conditions. These programs were adjusted according to the needs of the patients. At the time of discharge, the families of the patients were informed about the home exercise programs. During rehabilitation, radiotherapy and

chemotherapy were not applied to the patients.

Statistical analysis

Statistical analyses were carried out with SPSS for Windows version 15.0 (SPSS, Chicago, IL, USA). Continuous variables are presented as medians (interquartile range) and categorical variables as percentages. Differences between pre- and post-rehabilitation according to AIS score were assessed by marginal homogeneity test. The significance of the change in MMS, WeeFIM and KPS scores was evaluated by Wilcoxon signed rank test. The percentage change was calculated for the MMS, WeeFIM, and KPS scores. We evaluated whether there was a difference between risk factor groups (sex, diagnosis, tumor setting and level localization, recurrence, radiotherapy, chemotherapy, orthopedic problems, clinical symptoms pre-rehabilitation) according to the percentage changes in WeeFIM, MMS, and KPS scores *via* Mann-Whitney *U* or Kruskal Wallis tests. We used the Spearman correlation coefficient in order to determine a possible relationship between a numeric value (age, length of stay in neurosurgery unit, duration of rehabilitation) and the percentage change in MMS, WeeFIM and KPS score. The significance level was 0.05.

Author contributions: All authors participated in the study design, conduction, and evaluation, contributed to preparation of the manuscript, and approved the final version of this paper.

Conflicts of interest: None declared.

Peer review: This paper retrospectively analyzed the early rehabilitation impact on functional status of paediatric patients after spinal tumor surgical removal. There is very limited number of papers dealing with rehabilitation of paediatric oncological patients, so this paper is important as it fills the gap in the present-day knowledge.

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