

ORIGINAL ARTICLE

Current situation of central nervous system tumour survivors in a spanish hospital

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Adaptive behaviour;
Self-esteem;
Functional status;
Social factors;
Physical activity

Abstract

Introduction: Despite the increasing number of central nervous system (CNS) tumour survivors, long-term (LT) sequelae remain a substantial burden on their health through various life stages. The aim of our study was to evaluate late morbidity, health-related quality of life (HRQoL), self-esteem, functional status, adaptive behaviour, physical activity and social outcomes such as education, employment, relationship status and possession of a driver's license, in addition to the role of LT effects of radiotherapy (RTx) on these outcomes.

Methods: The study included 111 CNS tumour survivors with a minimum of 10 years of follow-up. The assessment of patients included a comprehensive clinical evaluation and psychological questionnaires: SF-36 for HRQoL and Rosenberg self-esteem scale. Functional status was assessed with the Lansky/Karnosky scales and adaptive behaviour with the Vineland-II scale. Patients also completed one questionnaire that assessed physical activity and another that assessed other social variables.

Results: The mean current age of CNS tumour survivors was 20 years (range, 12–34). The median time since diagnosis was 13 years (range, 10–26). The median age at diagnosis was 6 years (range, 1–18). The most prevalent LT sequelae were neurologic (55%), followed by neuropsychiatric (53.1%) and endocrine (49.5%) sequelae. Survivors treated with RTx (n = 52) had poorer outcomes in terms of adaptive behaviour, educational attainment and relationship status compared to those who did not receive RTx, but we did not find differences in quality of life or self-esteem scores between these groups.

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PALABRAS CLAVE

Largos supervivientes de cáncer;
 Tumores cerebrales;
 Efectos a largo plazo;
 Radioterapia;
 Calidad de vida relacionada con la salud;
 Comportamiento adaptativo;
 Autoestima;
 Estado funcional;
 Factores sociales;
 Actividad física

Conclusion: We found a high prevalence of long-term sequelae in CNS tumour survivors, especially in those who received RTx. In these patients, LT sequelae have a significant impact on functional status and social outcomes, but not on quality of life or self-esteem.

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Situación actual de supervivientes de tumores del sistema nervioso central en un hospital español

Resumen

Introducción: A pesar de la creciente población de supervivientes de tumores del sistema nervioso central en la infancia (TSNC), los efectos a largo plazo (ELP) siguen siendo una carga sustancial en las diferentes etapas de su vida. Este estudio buscó evaluar la morbilidad tardía y la calidad de vida relacionada con la salud (CVRS), así como la autoestima, el estado funcional, el comportamiento adaptativo, la actividad física y otras variables sociales como la educación, el empleo, la relación de pareja y posesión de permiso de conducir, además del impacto que podría tener el tratamiento con radioterapia (RT) en todos estos aspectos.

Métodos: Se incluyeron 111 Supervivientes de TSNC, con al menos 10 años de seguimiento. Se realizó una evaluación clínica y encuestas psicológicas: SF-36 para CVRS y Rosenberg para autoestima. Se estimó estado funcional con las escalas de Lansky/Karnosky y el comportamiento adaptativo con el Vineland-II, respondieron además una encuesta de actividad física y otra para las variables sociales.

Resultados: Los Supervivientes de TSNC, tuvieron una mediana de edad actual de 20 años (12–34), tiempo de seguimiento de 13 años (10–26) y edad al diagnóstico de 6 años (1–18). Los ELP más prevalentes fueron los neurológicos (55%), seguidos de los neuropsicológicos (53,1%) y endocrinológicos (49,5%). Los Supervivientes de TSNC tratados con radioterapia tuvieron peores resultados en la Escala de Comportamiento Adaptativo, nivel educativo y relación de pareja, pero no se encontraron diferencias en las puntuaciones de calidad de vida, ni en autoestima.

Conclusión: Los Supervivientes de TSNC presentan una alta prevalencia de efectos a largo plazo, especialmente aquellos que recibieron radioterapia. Estos ELP tienen un impacto significativo a nivel funcional y social, no así, en los resultados de calidad de vida y autoestima en este grupo.

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Introduction

Central nervous system (CNS) tumors are the most common solid tumors and the leading cause of death in childhood cancer.^{1,2} Advances in diagnosis and treatment have increased survival rates.^{3–5} According to the report of the Registry of Childhood Tumors of the Sociedad Española de Oncohematología Pediátrica (Spanish Society of Pediatric Oncohematology), the 5-year survival after diagnosis of a CNS tumor is 71%.⁶

Surviving a brain tumor is associated with a high risk of physical, neuropsychiatric and social late effects (LEs), especially in patients managed with radiotherapy (RTx).⁷ The most frequent LEs are neurologic, sensory and endocrine disorders, in addition to an increased cardiovascular risk. These disorders tend to be chronic and develop more than 5 years after treatment.^{8–14}

Late effects involving different systems can also contribute to neurocognitive changes ranging from mild learning difficulties to severe deficits, with a risk of progressive neurologic impairment years after treatment.^{15–17}

As follow-up progresses, CNS tumor survivors and their families need to redefine their priorities beyond the fear of recurrence to focus on LEs and their impact on social skills, independence and vocational goals.^{18–21} The literature on childhood brain tumor survivors describes a higher prevalence of psychological maladjustment and lower educational attainment, employment rates and frequency of partnered/romantic relationships. Psychosocial assessment is important in this population,^{22–24} as one of the main goals of follow-up in survivors should be to minimize the risk of exclusion.^{25–27}

Health-related quality of life (HRQoL) questionnaires are widely used in the follow-up of CNS tumor survivors, and it is surprising that, despite the multiple LEs that affect this group of patients, there are studies that do not find differences in HRQoL when comparing them to healthy controls.¹² On the other hand, a study found that childhood CNS tumor survivors overestimated their level of leadership-popularity and underestimated their isolation behaviors and victimization by peers.²⁸ This evinces the need to explore other aspects that are not addressed in these instruments, such

Table 1 Prevalence of late effects in CNS tumor survivors.

Late effects	n	%
Endocrine: hypothyroidism, growth hormone deficiency, diabetes insipidus, abnormalities in antidiuretic hormone secretion, overweight or obesity, adrenal insufficiency, abnormal glucose metabolism, other endocrine disorders related to central nervous system abnormalities	55	49.5%
Neurologic: seizures, cranial nerve disorders, dysarthria, headache, intracranial hemorrhage, movement disorders, cavernoma, autonomic dysfunction, cerebellar dysfunction, stroke, hydrosyringomyelia, peripheral motor or sensory neuropathy, intracranial hypertension, hydrocephalus, shunt malfunction, pseudomeningocele and other central nervous system abnormalities	61	55.0%
Neurocognitive: attention deficit, executive function deficit, fine motor dexterity deficit, memory deficit, processing speed deficit, other neurodevelopmental disorders	59	53.1%
Visual: anterior segment, retina or optic nerve abnormalities, reduced visual acuity, visual field deficit, blindness, strabismus, diplopia	41	36.9%
Auditory: hearing impairment	16	14.4%
Psychiatric/psychological: suicidal ideation, anxiety, depression, hyperactivity, oppositional disorder, post-traumatic stress disorder, sleep disturbances, other disturbances	13	11.7%

Late effects according to the categories of system-based chronic and late health events, applying the Common Terminology Criteria for Adverse Events (CTCAE) as modified in the St Jude LIFE study.

as adaptive behavior, including daily performance, independence and cultural expectations,²⁹ as well as other psychosocial variables, since they are a group of survivors with specific risks and whose social functioning could be impaired.^{27,29,30}

The aim of our study was to assess morbidity in a group of Spanish childhood brain tumor survivors beyond their HRQoL to cover aspects that have not been considered previously, such as adaptive behavior, self-esteem, physical activity as well as other social indicators such as independent living, education and relationship status, which may be important in the assessment of their functioning. We also sought to estimate the probable impact of radiotherapy on the physical and psychosocial areas under study.

Material and methods

We conducted a cross-sectional observational study in CNS survivors included in the childhood oncology database of our hospital.

The inclusion criteria were having undergone treatment between 1990 and 2010, age less than 18 years at diagnosis and active user status in the public health care system. We excluded patients currently in treatment for recurrence.

In the survivor registry, we identified 264 patients with a diagnosis of CNS tumor. Of this total, 221 were active in the health care system and we were able to locate 195, of whom 150 met the inclusion criteria and 111 agreed to participate in the study.

The study was approved by the ethics committee of the hospital and we obtained written informed consent from all the patients or, if they were under 18 years of age, from their parents.

All CNS tumor survivors were active patients at the long-term follow-up unit. We reviewed their health records to collect data on clinical variables including age, sex,

tumor type and location, age at diagnosis, predisposing genetic syndromes, treatment received, medical specialties involved in their follow-up, reported LEs, and duration of follow-up.

We analyzed LEs in these patients based on the information documented in medical reports and their detection in visits to the long-term follow-up unit. When a new LE was detected, survivors were referred to the appropriate specialist.

We classified LEs based on the involved organ or system applying the Common Terminology Criteria for Adverse Events (CTCAE) as modified in the St. Jude Lifetime Cohort Study,³¹ summarized in Table 1.

All study participants completed the following questionnaires:

- 1 36-Item Short Form Health Survey (SF-36) for assessment of HRQoL, which includes 8 subscales (physical functioning, role limitations [due to physical problems], bodily pain, general health, energy/fatigue, social functioning, role limitations [due to emotional problems] and mental health), with each item rated on a scale from 0 to 100.²⁸
- 2 Rosenberg self-esteem scale, which classifies self-esteem based on the score as low (0–15), normal (15–25) or high (25–40).³²
- 3 Lansky scale (in patients aged < 16 years) or Karnofsky scale (older patients) to assess functional/performance status.
- 4 Vineland Adaptive Behavior Scales, Second Edition (Vineland-II), administered through a personal interview with the survivor, also including a parent/legal guardian in patients aged less than 18 years and adult patients who authorized their presence. It yields scores in four domains (communication, daily living skills, socialization and motor skills) plus an adaptive behavior composite score. Each skill is rated on a scale from 0 to 2, all scores are age-standardized (100 ± 15) and, depending on the

Table 2 Clinical characteristics of CNS tumor survivors.

Characteristics of survivors (n = 111)	n	%
<i>Tumor location</i>		
Supratentorial	37	33.3%
Infratentorial	67	60.4%
Spinal	7	6.3%
<i>Type of CNS tumor</i>		
Low-grade glioma	57	51.4%
Medulloblastoma	18	16.2%
Ependymoma	6	5.4%
Tumor germinal	10	9.0%
Other high-grade tumors ^a	7	6.3%
Other low-grade tumors ^b	13	11.7%
<i>Treatment</i>		
Only surgery	47	42.3%
Surgery and radiation	14	12.6%
Surgery and chemotherapy	6	5.4%
Surgery + chemotherapy + radiation	33	29.7%
Surgery + chemotherapy + autologous transplant	5	4.5%
Surgery + chemotherapy + radiation + autologous transplant	6	5.5%
More than one surgery (including progression and relapse)	18	16.2%1
Current carrier of ventriculoperitoneal shunt	7	6.3%
<i>Radiation dose</i>		
None	58	52.2%
>50 Gy	36	32.4%
<50 Gy	17	15.3%
Tumor predisposition syndrome	10	9.0%
Second tumor	7	6.3%

^a Other high-grade tumors (n): glioblastoma multiforme (1), choroid plexus carcinoma (3) anaplastic ganglioglioma (1), primitive neuroectodermal tumor (2).

^b Other low-grade tumors (n): craniopharyngioma (6), meningioma (2), choroid plexus papilloma (3).

results, adaptive behavior is classified as high (>130), moderately high (115–129), adequate (86–114), moderately low (71–85) or low (<70).²⁹

- 5 Physical Activity Questionnaire for Adolescents (PAQ-A) or International Physical Activity Questionnaire-Short Form (IPAQ-SF), depending on the age of the participant. The first yields a score calculated as the mean score for its 9 items, each of which is rated on a 5-point scale, and the second assesses walking, moderate-intensity activity and vigorous-intensity activity, calculating the time spent on each activity (minutes) and its frequency (days) weekly and categorizes the level of physical activity as high, moderate or low.³³
- 6 Sociodemographic data form, which includes variables such as marital status, partnered/romantic relationship status, educational attainment, independent living and having an active driver's license.
- 7 In addition, a neurocognitive evaluation was only performed in underage patients who visited the pediatric neuropsychiatry clinic of the hospital with their parents.

Statistical analysis

We characterized the population of childhood CNS tumor survivors. We analyzed the distribution of the different variables with the Shapiro-Wilk test. We classified survivors based on whether they had received radiotherapy to com-

pare the LEs; calculating the prevalence ratio (PR) with the corresponding 95% confidence interval (CI) for each group and analyzing the differences in questionnaire results by means of the Wilcoxon-Mann-Whitney test, which we also used to compare follow-up duration based on the adaptive behavior scores.

The Spearman correlation coefficient was used to analyze the association between questionnaire scores.

We carried out a multiple linear regression analysis. The dependent variable was adaptive behavior and the independent variables the age at diagnosis, dose of radiation and time elapsed since diagnosis. The diagnosis of brain tumor was not included in the analysis, since it was strongly correlated to the treatment variable and the latter was associated more frequently with LEs.

The size of the subset of patients who underwent neuropsychiatric assessment was too small to be representative for the purpose of hypothesis testing.

We defined statistical significance as a *P* value of less than .05 for all tests. All the statistical analyses were performed with the software IBM SPSS, version 26 (SPSS Inc; Chicago, IL, USA).

Results

The study included 111 childhood CNS tumor survivors (70 male). The median current age was 20 years (range, 12–34),

Table 3 Comparison of SF-36 health-related quality of life scores in CNS tumor survivors who received radiotherapy versus those who did not.

HRQoL score	RTx group, mean \pm SD	No RTx group, mean \pm SD	P
Physical functioning	86.97 \pm 9.15	89.70 \pm 7.56	.41
Role limitation (physical)	83.13 \pm 8.20	86.66 \pm 9.16	.49
Bodily pain	81.84 \pm 11.77	80.08 \pm 9.16	.95
General Health	70.13 \pm 18.83	73.50 \pm 20.26	.45
Energy/fatigue	69.21 \pm 17.61	70.50 \pm 20.48	.47
Social functioning	83.28 \pm 17.61	84.75 \pm 14.31	.77
Mental health	76.68 \pm 17.43	78.86 \pm 11.41	.32

HRQoL, health-related quality of life; RTx, radiotherapy; SD, standard deviation.

the median time elapsed from diagnosis was 13 years (range, 10–26) and the median age at diagnosis was 6 years (range, 1–18). [Table 2](#) summarizes the clinical characteristics of the CNS tumor survivors, including the diagnosis, tumor location and received treatment.

Late effects

The prevalence distribution of LEs in CNS tumor survivors was: neurologic (55.0%), neuropsychiatric (53.1%), endocrine (49.5%), ocular/visual (36.9%) and auditory/hearing (14.4%). [Table 1](#) presents the most frequent diagnoses by LE category according to the St. Jude LIFE classification based on the National Cancer Institute's Common Terminology Criteria for Adverse Events (CTCAE).

We compared CNS tumor survivors treated with radiotherapy (RTx group, n = 53) to those who did not receive this treatment (no RTx group, n = 58). In the RTx group we found a significantly higher prevalence of endocrine (PR, 4.21; 95% CI, 2.43–8.89), neurologic (PR, 1.89; 95% CI, 1.34–2.66), neurocognitive (PR, 1.70; 95% CI, 1.12–2.57), visual/ocular (PR, 2.69; 95% CI, 1.59–4.53) and auditory (PR, 2.17; 95% CI, 1.78–2.64) late effects.

Health-related quality of life

We compared the impact of radiotherapy on HRQoL and found no statistically significant differences. [Table 3](#) compares the results obtained in the different instruments in patients who received RTx versus patients who did not.

Self-esteem

The score on the Rosenberg scale was 30.2 (SD, 2.1) in the RTx group compared to 31.3 (SD, 3.3) in survivors who did not receive RTx ($P = .61$).

The scores were below normal in 5.6% of patients and in the high range in 43%. We found no significant differences when we compared the scores in the two treatment groups.

We found a positive and moderately strong correlation between the total scores in the quality of life questionnaire and the self-esteem questionnaire (Spearman rho = 0.462; $P = .001$) ([Fig. 1](#)).

Adaptive behavior

In the comparison of adaptive behavior scores (for each domain and overall composite score) in CNS tumor survivors

based on the treatment history, we found that those who received radiotherapy scored significantly lower in communication, daily living skills and socialization ([Table 4](#)).

We used multiple linear regression to assess the predictive value of the independent variables (age at diagnosis, radiotherapy treatment and number of LEs) in relation to the Vineland-II composite score. The general linear model explained 52.2% ($R^2 = 0.522$) of the variance in the composite score ($P < .01$; $F_{3,44} = 16$; $\beta = 2.63$). Each additional year of age at diagnosis was associated with an increase in the dependent variable of the Vineland composite score ($\beta = 1.00$), in contrast to increases in the dose of radiation ($\beta = -0.26$) or in the number of LEs ($\beta = -5.35$), which predicted poorer scores in the Vineland-II ($P < .01$).

Based on the composite adaptive behavior score, we classified survivors into 2 groups: "adequate to moderately high" and "moderately low to low". Patients with poorer adaptive behavior scores had a significantly longer duration of follow-up in years ($P = .04$) ([Fig. 2](#)).

The mean score in the Lansky or Karnofsky scale used for assessment of functional status was 80.8 (SD, 12.1) in the RTx group compared to 89.6 (SD, 8.5) in survivors who did not receive RTx ($P = .01$).

There was a strong positive correlation between functional status and adaptive behavior scores (Spearman rho = 0.796) ([Fig. 3](#)).

Sociodemographic variables

Sixty-three percent of CNS tumor survivors were older than 18 years at the time of the study and 26% were older than 25 years. Of all the adult survivors, only 13.2% lived independently, without a parent or caregiver at home (RTx group, 12.5% vs No RTx group, 13.8%; $P = .83$). When it came to educational attainment, 21.7% of CNS tumor survivors had started secondary education or university (RTx group, 13.8% vs No RTx group, 31.3%; $P = .014$); 17.9% did not currently have a romantic partner (RTx group, 12.1% vs No RTx group, 18.5%; $P = .035$) and 30% reported never having had a romantic partner (RTx group, 29.3% vs No RTx group, 20.3%; $P = .04$). Only 17.3% were employed (RTx group, 15.1% vs No RTx group, 17.2%; $P = .75$) and 21.7% had obtained a driver's license (RTx group, 20% vs No RTx group, 23%; $P = .83$).

The mean physical activity score in survivors aged less than 18 years (PAQ-A) was 1.4 (SD, 0.64). Based on the results of the IPAQ-SF, survivors aged more than 18 years engaged in vigorous-intensity physical activity for a mean of 26 min a

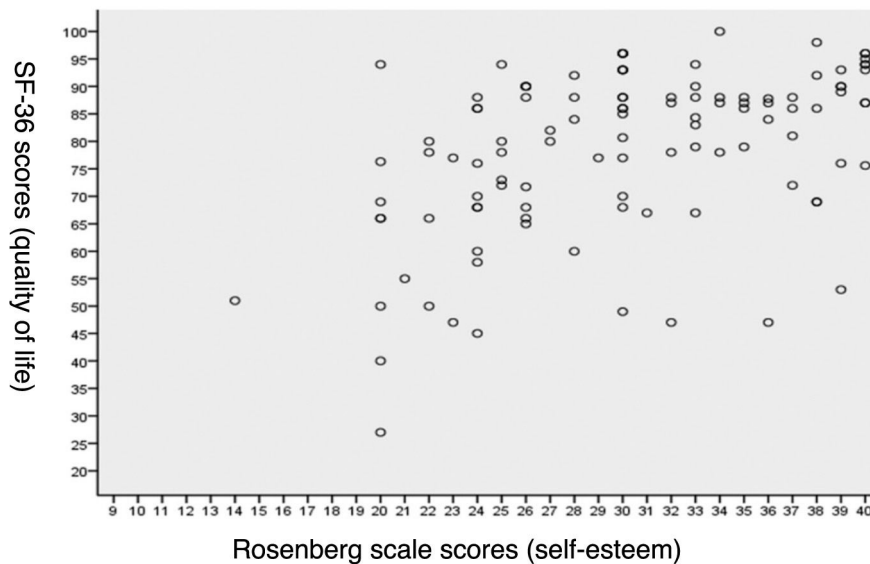


Figure 1 Association between scores in the self-esteem scale and the health-related quality of life survey. We found a positive and moderate-to-strong correlation between the self-esteem and quality of life scores ($\rho = 0.462$; $P = .001$). Rho, Spearman correlation coefficient.

Table 4 Comparison of scores on the Vineland-II scales (domain and composite scores) in CNS tumor survivors who received radiotherapy versus those who did not.

Vineland-II scores	Overall CNS tumor survivors, mean \pm SD (range)	RTx group mean \pm SD	No RTx group mean \pm SD	<i>P</i>
Communication	98.7 \pm 18.4 (35–120)	93.1 \pm 22.1	106.52 \pm 8.2	.004
Daily living skills	96.8 \pm 21.3 (30–130)	89.8 \pm 21.4	107.73 \pm 16.7	<.001
Socialization	96.7 \pm 17.2 (32–130)	93.5 \pm 21.3	101.71 \pm 14.6	.024
Motor skills	96.9 \pm 18.4 (58–130)	95.2 \pm 18.7	100.48 \pm 17.8	.102
Composite score	97.9 \pm 18.4 (36–118)	92.8 \pm 18.9	104.2 \pm 12.5	.001

CNS, central nervous system; RTx, radiotherapy; SD, standard deviation.

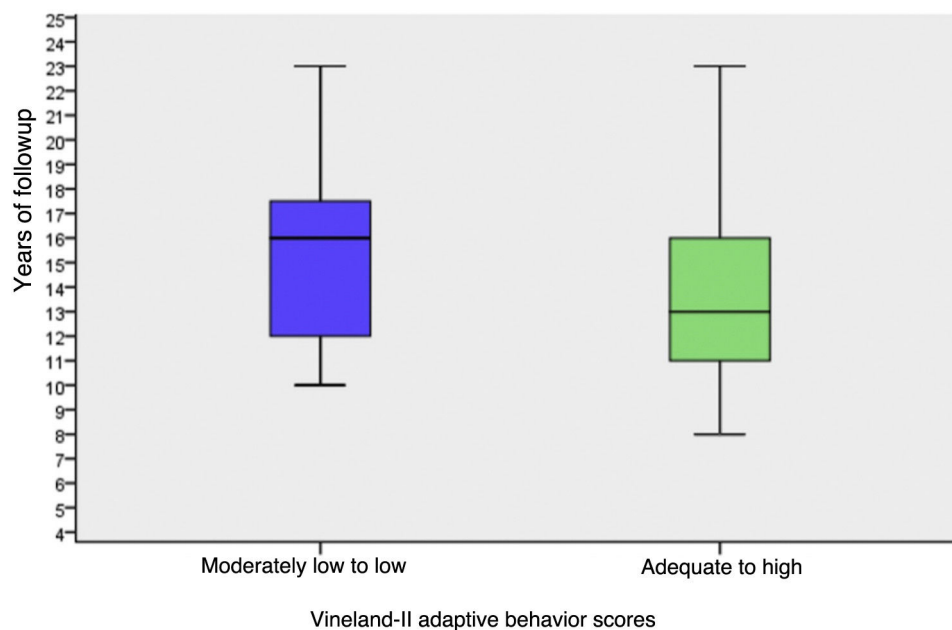


Figure 2 Comparison of CNS tumor survivors with moderately low to low versus adequate to high composite adaptive behavior scores based on duration of follow-up. The Wilcoxon rank test found a significant difference in the duration of follow-up between the two groups. $P = .04$.

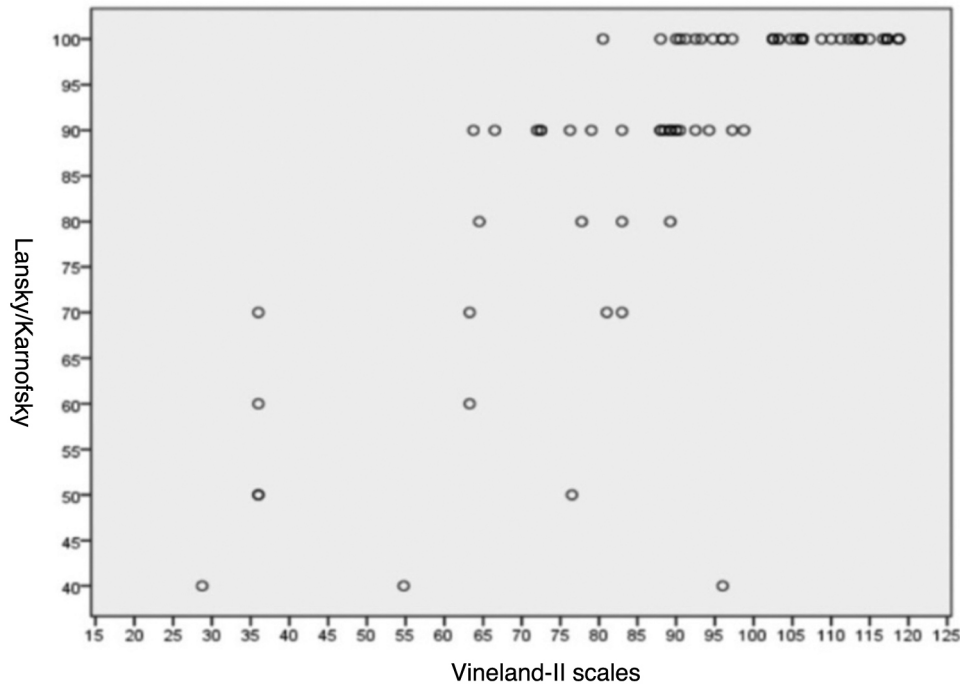


Figure 3 Association between the scores in the Vineland-II adaptive behavior scales and the Lansky/Karnofsky performance scales. We found a strong positive correlation between the functional status and adaptive behavior scores ($\rho = 0.796$; $P = .001$). Rho: Spearman correlation coefficient.

week (SD, 68), moderate-intensity activity for 420 min (SD, 232) and low-intensity activity for 2450 min (SD, 1230).

Thus, of the total CNS tumor survivors, 13% had a moderate level of physical activity, and only 1.8% had a high level of physical activity.

The neuropsychiatric evaluation was only completed in 26% of the sample, which corresponded to 72% of the patients evaluated at the hospital because they were underage. Seventy-nine percent of patients had overall scores at the 50th percentile or higher, 13% exhibited mild impairment and 7% moderate to severe impairment.

Discussion

The distribution of the diagnoses and locations of CNS tumors in this sample of survivors was consistent with the previous literature.⁴⁻⁶

For the purpose of analysis, we divided the sample of survivors based on whether or not they had been irradiated, since radiotherapy, in addition to being a known predictor of poorer outcomes and LEs is strongly associated with the diagnosis of more aggressive and less frequent tumors, which we grouped together in order to obtain a group size comparable to the size of the group of patients who did not receive radiotherapy, who predominantly had low-grade tumors managed with surgery alone. This stratification allowed us to analyze differences in questionnaire scores to assess the potential impact of radiotherapy.

The most frequent LEs were neurologic and cognitive, followed by endocrine and neurosensory LEs. We found greater morbidity in patients managed with radiotherapy, as described in previous studies.³⁴⁻³⁷ However, we did not

find significant differences in HRQoL and self-esteem scores when we compared the two groups.

Although HRQoL is important in the cancer survivor population, most questionnaires assessing this aspect do not cover aspects such as cognitive skills, family dynamics, social interactions, partnered/romantic relationships, educational attainment or employment.³⁸ Similarly, the use of instruments for assessment of self-esteem has been questioned due to known limitations, especially in groups with potential cognitive impairment,³² and another factor that could be at play is the underdiagnosis of LEs in this group of patients.

The observed positive and moderately strong correlation between HRQoL scores and self-esteem scores in our study suggests that these indicators may be more variable and subjective compared to others, like adaptive behavior and functional status, in CNS tumor survivors.

On the other hand, we found significant differences in adaptive behavior and functional status scores when we compared CNS tumor survivors based on whether or not they had been managed with radiotherapy. In the bivariate analysis, we also found a strong correlation between the scores in these two scales.

Adaptive behavior is a relevant indicator in this subset of survivors. A study in patients treated with conformal radiation therapy found significantly lower adaptive functioning scores compared to controls, and this treatment was also correlated to poorer neurocognitive scores.²⁹

In the multiple linear regression analysis, the adaptive behavior scores worsened with decreasing age at treatment, especially below age 6 years, with increasing radiation dose and with increasing duration of follow-up. Furthermore, when we compared the groups with "adequate to mod-

erately high” versus “moderately low to low” adaptive behavior scores, we found a significantly longer median duration (in years) of follow-up in the latter group. A possible explanation is that survivors in the group with poorer scores may not be able to keep pace with their same-age peers so that the developmental gap between them would widen over time, chiefly due to progressive neurologic deterioration.^{15,16}

With respect to social variables, the percentage of adult survivors who had enrolled in university or were employed was remarkably low. In the group of CNS tumor survivors who had received radiotherapy, there was a higher percentage who had dropped out after completing primary education and a higher percentage who had never had a romantic partner.

Promoting physical activity is important in CNS tumor survivors, who are at increased risk in several areas in association with LEs. In addition, there is evidence that exercise is associated with better neurocognitive outcomes and better social integration in CNS tumor survivors.^{39,40} Eighty-five percent of survivors in our study reported low levels of physical activity in the questionnaires, compared to the results found in the Spanish validation study of the international physical activity questionnaire.³³

Among the limitations of our study, we ought to mention that, since our hospital only offers neurocognitive evaluation in the pediatric age group, the size of the group that had undergone this assessment was insufficient for analysis of this aspect. The design and sample of the study did not allow us to make comparative analyses based on the type or location of the tumor, and, since the duration of follow-up was longer than 10 years, all survivors treated with radiotherapy had received conventional irradiation, with larger doses and fields than those used at present, and new treatment modalities such as proton therapy were not represented.

We believe that a more holistic approach to the follow-up of CNS tumor survivors, involving them actively along with their families in the detection and prevention of LEs, in addition to a more thorough assessment of functional performance and psychosocial functioning, could be a good first step to improve their transition to adult care under different specialists.

Conclusion

The prevalence of LEs in pediatric CNS tumor survivors is high, and even higher in those managed with radiotherapy. The history of radiotherapy was not associated with the self-reported HRQoL or self-esteem, but it was associated with adaptive behavior, the level of functioning and certain social variables, such as educational attainment and partnered/romantic relationship status.

Additional studies in larger samples and with more thorough assessments are required to improve our knowledge of long-term outcomes and follow-up in pediatric CNS tumor survivors.

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Declaration of competing interest

The authors have no conflicts of interest to declare in relation to the research, authorship or publication of this article.

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