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**Long-term burden of informal caregiver 7-years after severe childhood Traumatic
Brain Injury in the Traumatisme Grave de L'Enfant (TGE) study**

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Abstract

Objective: To investigate reported burden by the Primary Family Caregiver (PFC) 7-years after severe pediatric traumatic brain injury in the TGE (*Traumatisme Grave de l'Enfant*) longitudinal study.

Methods: Subjective burden was estimated with the Zarit Burden Inventory (ZBI) in 36 PFC (parents), who rated their own health status (Medical Outcome Study Short Form-12), family functioning and their child's level of care and needs (Pediatric/Adult Care And Needs Scale [PCANS/CANS]). Data collection included: child and PFC sociodemographic characteristics, injury-related factors, "objective" (*e.g.*, overall level of disability: Glasgow Outcome Scale–Extended, GOS-E/GOS-E-Peds) and "subjective" outcomes (*e.g.*, participation, behavior, executive functions, quality of life and fatigue).

Results: 25% of PFC reported mild-moderate burden, and 19% moderate-severe burden. Higher burden correlated with worse outcomes in all "subjective" PFC-rated outcomes, and with self-reported participation. The ZBI correlated strongly with CANS/PCANS and GOS-E/GOS-E-Peds. Overall level of disability and PFC-reported executive functioning explained 62% of the ZBI variance. For equal levels of disability, burden was higher when PFC reported a "negative" picture of their child.

Conclusion: Significant PFC-reported burden 7-years post-injury was associated with overall disability and "subjective" PFC-rated outcomes. Factors influencing parental burden in the long term should be identified and psychological support implemented over time.

Key words: informal caregiver, primary family caregiver, severe traumatic brain injury, child, multidimensional burden, prospective cohort study.

Long-term burden of informal caregiver 7-years after severe childhood Traumatic Brain Injury in the Traumatisme Grave de L'Enfant (TGE) study

Childhood Traumatic Brain Injury (TBI) is the main cause of mortality and long-standing disability in children, constituting a major public health issue (1). After severe TBI, common outcomes encompass impairments in cognitive, behavioral and emotion regulation (2–4), which negatively impact subsequent autonomy, academic and professional achievement (5).

Family members of children with multiple disabilities following severe TBI often experience health problems and limitations of other activities due to the burden of care and financial challenges (6). A limited number of studies have focused on the burden experienced by the child's informal caregivers during the sub-acute and chronic phases after pediatric TBI. A literature review (7) underlines a trend towards more dysfunction in families whose child had a severe TBI compared to families with a child who had a moderate TBI or orthopedic injury. Stancin et al. (8) compared the burden on informal caregivers of young children with orthopedic injury (*i.e.* patients with physical impairments but no brain injury) with the burden on informal caregivers of young children with TBI. Informal caregivers dealing with cognitive impairment issues reported a higher overall burden. Furthermore, informal caregivers of children with severe TBI reported poorer relationships with spouses and siblings, as well as higher levels of depression and global distress. Additionally, poorer psychological functioning of caregivers has been associated with the presence of behavioral problems during adolescence in individuals who sustained pediatric TBI (9). In contrast, Wade et al. (10) reported no statistical differences in “marital distress” between parents of children with TBI and parents of children with orthopedic injuries during the first year post-injury. Aitken et al. (11) investigated parental burden at 3 and 12 months after pediatric TBI.

Parental burden was associated with the severity of cognitive impairment in the child, as well as unmet healthcare needs according to informal caregiver's perception. In a 5-year prospective study, Mangeot et al. (12) reported a relationship between greater executive dysfunctions in the child and higher parental stress and burden. Some studies found an age-at-injury effect on informal caregivers' burden (8,13). For instance, informal caregivers of children aged 5 to 6 years at the time of injury appear to experience significantly higher levels of both injury-related burden and distress than those caring for children aged 3-4 years at injury (8).

The enduring negative impact of TBI on family functioning and caregivers is chronic and can may persist over many years (14). While most studies have focused on informal caregivers' burden within the first-year post-injury, the long-term impact of TBI on family functioning and informal caregivers' burden has been overlooked. In addition, there has been a lack of differentiation between objective and subjective factors influencing reported informal caregiver burden. Regarding the burden of informal caregivers for individuals who sustained TBI in adulthood, a review (15) described consistently high levels of distress among informal caregivers, regardless of caregiver type (*i.e.* spouses *vs.* parents). The major concern among informal caregivers of adults who sustained TBI revolves around determining who will provide care when mothers are no longer present or able (13). Previous longitudinal studies and literature reviews, focusing on individuals aged ≥ 15 years at the time of injury with follow-up ranging from 1 to 10 years (16,17), have evidenced a decrease in subjective informal caregiver burden over time. These findings are probably the result from improvements in both patients' autonomy (18) and informal caregivers' coping capacity (19) over time. However, it is noteworthy that contrasting results (20) have also been reported.

The present study is part of a 7-year longitudinal prospective longitudinal cohort study (see Methods). The primary aim of this study was to evaluate the subjective level of burden

reported by the primary family caregiver 7 years after severe pediatric TBI, using the Zarit Burden interview (ZBI) scale (21). Secondary aims included examining the relationship between the reported burden and: (i) sociodemographic characteristics of the patient and informal caregiver (*i.e.* age, gender, socioeconomic status); (ii) injury-related factors, such as age at injury, indices of injury severity (Glasgow Coma Scale (GCS) score, Injury Severity Score (ISS) and length of coma); (iii) family-related factors, including family functioning, parental mental health status, and level of care and needs perceived by the primary family caregiver (assessed using age-appropriate forms of the Care And Needs Scale (22,23); (iv) concurrent outcomes at 7-years post-injury, distinguishing between (a) “objective” (Glasgow Outcome Scale - Extended (GOS-E) (24) and GOS-E-Pediatric version (25), intellectual ability, presence of motor deficits, educational/professional situation) and (b) “subjective” outcomes (questionnaires completed by the primary family caregiver and/or the patient) describing the patient’s executive functioning, behavior, participation, quality of life and fatigue.

We hypothesized: (a) elevated levels of caregiver burden; (b) strong associations between reported burden with objectively-assessed functional outcomes; (c) some association between reported burden and questionnaire-based assessments of the patient’s status, especially with executive and behavioral difficulties; (c) strong correlations between burden and the Care and Needs scales; (d) a possible association of burden with primary family caregiver’s physical and mental health, as well as patient/primary family caregiver socio-demographic characteristics; (e) a significant association of burden with family functioning.

Material and methods

Study design

This study draws from the *Traumatisme Grave de l’Enfant* (TGE) (*i.e.* severe childhood traumatic brain injury) prospective longitudinal cohort of children (<16 years),

consecutively hospitalized in a single intensive care unit following accidental severe TBI (for further details on enrolment procedure, initial severity and outcomes, see 26–29). From the 81 children included initially, 16 died during acute care, leaving 65 for subsequent follow-up assessments.

Participants

At 7-years post-injury, 26 of the 65 patients were lost to follow-up, or their families chose not to participate, and 3 participants did not complete the main outcome measures. The 36 patients assessed 7-years post-injury, along with their primary caregivers, did not differ from those lost to follow-up or with incomplete data regarding sociodemographic background, injury severity characteristics, and follow-up outcomes at 3, 12 and 24 months post-injury (27).

Data collection

1. Primary Family Caregiver

1.1. Socio-demographic and family characteristics

For the present study, we focused on primary informal (family) caregiver of the patients, defined as the individual primarily responsible for care during the years post-injury or involved in day-to-day decision making (30). We collected data on sociodemographic (age and gender) and family characteristics of the primary family caregiver, including the degree of kinship, co-residency, the presence of other children in the family, other family members requiring caregiving, parental education level (*medium/high*: at least one parent graduated from high school or *low*: none of the parents reached high school graduation), and professional activity (active or non-active).

1.2. Zarit Burden Inventory (ZBI)

Subjective burden was assessed using the Zarit Burden Inventory (ZBI) (21), which contains 22 multidimensional questions rated as 0=Never, 1=Rarely, 2=Sometimes, 3=Quite

frequently, 4=Nearly always. The total score (possible range 0 to 88) represents the sum of all items. Proposed clinical cut-offs are as follows: mild (range: 0–20), mild to moderate (21–40), moderate to severe (41–60), and severe burden (61–88) (31). Previous reports have indicated the usefulness of the Zarit Burden Interview in the assessment of burden in caregivers of individuals acquired brain injuries (32).

1.3. Physical and Mental Health of the Primary Family Caregiver (self-report)

We used self-report versions of the 12-Item Short-Form Health Survey (SF-12) (33) to evaluate the health status of the primary family caregiver. We calculated two summary scores: Physical (PCS) and Mental Component Summary (MCS). These scores were standardized to reflect a general population mean of 50 (SD=10), with higher scores indicating better health.

1.4. Family functioning

The primary family caregiver completed French versions of the 12-item Family Assessment Device (FAD) (34). Higher scores (range 1 to 4) indicate poorer family functioning.

1.5. Care and Needs Scales (CANS/PCANS) according to the primary family caregiver

We used the Pediatric and Adult Care and Needs Scales (PCANS/CANS) for ages <16 (22) and ≥ 16 years (23) to categorize children with a “high level of needs” based on all scores above the highest value observed in a matched control group (see 39 for further details).

2. Participants who sustained a severe TBI

2.1. Sociodemographic data

We retrieved information on participant’s age (at the time of injury and at assessment), gender, and pre-injury education status (regular education or assisted/delayed).

2.2. Initial injury severity

We assessed initial injury severity during the acute phase in the ICU with the Pediatric GCS score (36). The minimal GCS score within the first 24 hours was recorded to account for

possible oscillations in this score. Additionally, we used the Pediatric Trauma Score (PTS) (37), the Injury Severity Score (ISS) (38), and the length of coma (in days).

2.3. Seven-years post-injury assessment (concurrent outcomes)

2.3.1 Overall level of disability assessed with the Glasgow Outcome Scale-Extended (GOS-E) (24) and its Pediatric version (GOS-E-Peds) (25): Upper (=1) or Lower (=2) Good Recovery, Upper (=3) or Lower (=4) Moderate Disability, Upper (=5) or Lower (=6) Severe Disability, 7=Vegetative State and 8=Death.

2.3.1. Presence of motor impairments: Presence or absence of motor deficits (*e.g.*, hemiplegia/hemiparesis) and/or signs of cerebellar dysfunction assessed through neurological examination.

2.3.2. Ongoing education: Mainstream (enrolment in general education classrooms, with or without support and/or repeated year), or special education.

2.3.3. Intellectual ability assessed with the Wechsler Intelligence Scale for Children-IV (WISC-IV) (39) and Wechsler Adult Intelligence Scale-IV (WAIS-IV) (40): Full Scale Intellectual Quotient (FSIQ, M=100, SD=15).

2.3.4. Executive Functioning evaluated with the Behavior Rating Inventory of Executive Function (BRIEF) for ages <18 years (41) and Adult-BRIEF for ages ≥18 years (42): Higher *T*-Scores M=50, SD=10) indicate worse executive functioning.

2.3.5. Behavior measured with the Child or Adult Behavior Checklist (CBCL, ABCL) for ages 6 to 18 years and/or Youth or Adult Self Report (YSR, ASR) from 11 years) (43,44): Higher *T*-Scores (M=50, SD=10) express more behavior problems.

2.3.6. Health-related Quality of life assessed with the Pediatric Quality of Life Inventory (PedsQL) for ages 2-18 and 18-25 years (48, 49): Higher total scores (range 0-100) indicate better-reported HRQoL.

2.3.7. *Fatigue* measured with the Multidimensional Fatigue Scale (MFS) for ages 2-18 and 18-25 years (50, 51)]: Higher total scores (range 0-100) suggest lower fatigue levels.

2.3.8. *Participation* assessed with the Child and Adolescent Scale of Participation (CASP) (47,48): Higher total scores (range 0-100) indicate better participation levels.

Procedure

The present work follows the guidelines outlined in the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement (49). This study was approved by the *Comité de Protection de Personnes d'Île de France VI* [CPP IDF VI] and by the Consultative Committee for Treatment of Health Research Information. Parents of children and adult participants who agreed to participate in the study provided written informed consent. Assessments of participants with TBI 7-years post-injury occurred during two sessions, encompassing medical and neuropsychological assessments (for further details, see 51–53).

Statistical analyses

Statistical analyses focused on reported burden (ZBI) as the main continuous variable to explain. Univariate analyses (Student's *t* and Pearson correlations) were first performed to identify variables significantly associated (α error set at 5%) with the ZBI. Additionally, we examined the correlation matrix (and partial correlations after adjusting for overall disability: GOS-E) between the main explanatory variables, which evidenced strong correlations among all questionnaires completed by the caregiver. Multivariate regressions aimed at explaining the variability of the ZBI included an examination of the role of initial injury severity variables when considering the GOS-E, and a backward selection of the concurrent outcomes (GOS-E and caregiver-rated questionnaire-based variables) assessed 7-years post-injury.

Results

Caregiver outcomes (ZBI, SF-12 and FAD) and level of care and needs (CANS/PCANS) according to the primary family caregiver (Table 1)

The mean age of primary family caregivers was 45.4 years (SD=7.9; range=29-64). All but one were a parent (72% mother, 25% father; 1 spouse), with 78% residing in the same household and 58% being active workers (*i.e.* engaged in current professional paid activity on the job market). The mean ZBI score was 21.14 (SD=17.30; range 0-54). Twenty primary family caregivers (56%) reported mild burden, nine (25%) mild to moderate, and seven (19%) moderate to severe burden. Health status, as assessed by the SF-12 questionnaire, reached a mean value of 46.67 (SD=10.25; range 25.29–62.12; %<1.5 SD=14%) for the physical component and 43.56 (SD=11.20; range 17.15–59.38; %<1.5 SD=25%) for the mental component, the latter being significantly different from the mean (SD) expected value of 50 (10) in the general population ($t_{(35)}=3.45$, $p=0.0015$). Primary family caregivers reported significantly high levels of needs (PCANS/CANS) for 35% of the participants aged <16 years and for 69% of the participants ≥ 16 years. The mean score in the Family Assessment Device (FAD) was 1.6 (SD=0.8; range 1-3; possible range: 1=best functioning to 4=worse functioning), indicating that family functioning was generally reported as good.

Participants with severe childhood TBI (Table 1)

Table 1 presents the main sociodemographic characteristics and initial injury severity markers of the study participants. The sample included 36 patients, with 14 (39%) girls. The mean age at injury was 7.53 years (range 3 months to 14.67 years), mean age at assessment was 15.31 years (range 7.42 to 22.67 years), and mean time since injury was 7.78 years (range 6 – 9). The mean GCS score was 5.97 (range 3–8; median 6), mean ISS score 27.58 (range 4–50; median 29), and mean length of coma 6.52 days (range 1–22 days; median 5 days)].

The GOS-E and GOS-E-Peds scores 7-years post-injury were distributed as follows: 20 participants (56%) had a “good recovery” status (*i.e.* GOSE 7 to 8), 8 (22%) had “moderate disability” (*i.e.* GOS-E 5 to 6), and 8 (22%) had “severe disability” (*i.e.* GOS-E 3 to 4). Twelve participants (33%) attended special education institutions. The mean FSIQ fell 1 SD below expected values, but showed remarkable variability (range 40-129), with 4 participants (11%) scoring below 70. Table 1 also presents a summary of scores obtained from the five self- and proxy-reported questionnaires. Caregivers reported difficulties within the clinical range for executive functioning (44%), behavior problems (42%), participation (50%), quality of life (36%), and fatigue (64%). Participants’ self-reports indicated similar mean scores and close proportions of low quality of life (31%) and increased fatigue levels (53%), but lower proportions of executive functioning difficulties (27%), behavior problems (26%) and reduced participation (19%).

Details of reported burden (item sorting of the ZBI)

The rank order of the 22 items from the ZBI completed by the primary family caregiver indicated that the most frequent concerns were related to the patient’s future, feeling that the patient is dependent upon the primary caregiver, difficulties in meeting responsibilities for family or work, feeling that the patient asks for more help than he/she needs, and feeling of dissatisfaction with the job of care the primary caregiver provides (see supplementary material Table s1). On the other hand, very few caregivers reported feeling uncomfortable about having friends over because of the patient or wishing to leave the care of the patient to someone else.

Correlations of reported burden with primary family caregiver’s/family characteristics, health status, level of care needs, and family functioning (Table 2)

The results of univariate analyses indicated that higher caregiver-reported perceived burden was associated with the presence of other family members in need of caregiving, as well as with an active professional activity of the primary family caregiver. Burden was also associated with poorer mental health status (SF12-MCS) of the caregiver, and with higher care and needs' levels for patients aged ≥ 16 years (according to the CANS) (see supplementary material Figure s1 and s2). The remaining associations of the ZBI with sociodemographic and family characteristics (including the gender of the respondent), the SF12-PCS, the PCANS, and family functioning (FAD) were not significant.

Associations of levels of burden with initial injury severity and 7-year post-injury concurrent outcomes (Table 2)

Regarding objective outcomes, higher reports of caregiver burden were significantly associated with markers of higher injury severity (higher ISS, longer length of coma), and with higher (worse) disability scores (GOS-E/GOS-E-Peds) (see Figure 1). When accounting for the effect of the GOS-E, the association of the ISS and length of coma were no longer significant. The associations of caregiver burden with motor impairments, FSIQ and type of ongoing education did not reach statistical significance.

Regarding questionnaire-reported outcomes, higher levels of caregiver burden were associated with caregiver reports of worse executive functioning, behavioral problems, participation, quality of life, and fatigue of the patient ($p < 0.001$ in all cases), whereas the association with patients' self-reports were not significant, except for self-reported participation levels ($p = 0.05$).

Correlations and partial correlations (GOS-E-adjusted) between burden and Primary Family Caregiver's reports of mental health and concurrent outcomes (Table 3)

Primary family caregiver mental health status (SF12-MCS) and questionnaires completed by the primary family caregiver (addressing the patient's executive functions,

behavior, participation, quality of life and fatigue) remained significantly associated with burden after overall disability (GOS-E) was considered. In addition, partial correlations (GOS-E adjusted) between the five questionnaires remained significant. Caregiver-reported everyday executive functioning, as assessed with the BRIEF, showed the strongest association with burden levels. All caregivers whose child's mean score in the BRIEF fell within the clinical range reported moderate to severe burden levels (see supplementary material Figure s3).

Regression analysis (Table 4)

The regression model, using the GOS-E, caregiver's mental health status, and the five questionnaires completed by the primary family caregiver as explanatory variables of perceived burden 7-years post-injury, indicated that none of the variables remained significant when all the other variables were partialled out. A backwards selection retained two variables: overall disability and executive functioning, which together explained 62% of the variance of the perceived burden 7-years post-injury.

Discussion

The results of this 7-year longitudinal prospective study can be summarized as follows: almost half of the primary family caregivers reported mild to moderate or moderate to severe burden levels (measured with the ZBI), 7-years after their child's severe TBI, with a mean burden level in the low range of "mild-to-moderate burden". Regarding factors associated with elevated burden levels, among the "objective" factors (situation- and performance-based variables, clinical ratings), only the overall level of disability (GOS-E) was significantly correlated with burden. On the other hand, elevated caregiver burden levels were associated with scores in the clinical range (*i.e.*, above the proposed clinical cut-off for each instrument) in all questionnaires assessing "subjective" patient outcomes completed by

the primary family caregiver. Among self-reported questionnaires, only the patient's self-reported participation showed a moderate correlation with caregiver-reported burden, with lower levels of patient participation associated with higher levels of caregiver burden.

Caring for a child with cognitive and physical impairments resulting from pediatric TBI is a stressful and challenging task, often becoming an all-encompassing activity and leading to significant changes in the informal caregivers' life style (53).

Compared to previous similar studies, the TGE cohort included a relatively large group of participants, exclusively focusing on children and adolescents with severe TBI followed up over 7 years. Studying a group of participants who have been fulfilling the role of primary family caregiver for several years provides a privileged perspective on the consequences of caregiving for a disabled child. Being so distant from the injury allows for a thorough analysis of the long-term burden, addressing both emotional concerns and disruptions in daily life activities, while minimizing the role of the acute initial stress and its impact on family members.

We assessed parental burden using the Zarit Caregiver Burden Interview, one of the most widely used measures of caregiver burden, considered as a unidimensional measure in the majority of previous studies (54). A descriptive analysis of the ZBI scores indicated that 25% of the primary family caregivers reported mild to moderate burden, and 20% indicated moderate to severe burden (17). Our study confirms a trend, already described in previous studies with shorter follow-up (7), for a higher stress level in families whose child had severe cognitive disability (*i.e.* severe TBI vs. moderate TBI vs. orthopedic injury). In accordance with previous reports, we found a correlation between the level of executive functions impairment and the level of reported burden (11,12,55). The level of overall disability (GOS-E) and of caregiver-reported everyday life executive functioning explained together 62% of the variance of the perceived primary family caregiver's burden 7-years post-injury. In

contrast, intellectual ability, although negatively correlated to the level of burden, as expected, did not reach significance, nor did the presence of motor deficits. Similar findings were reported after severe TBI in adolescence or adulthood (≥ 15 years), with overall disability and everyday executive functions impairments found to be independent predictors of caregiver burden 1-year post-injury (16). In children with cerebral palsy, a negative correlation has been reported between primary family caregivers' burden and the child's motor and communication skills (56).

Interestingly, all questionnaire-based scores completed by the same informant (self or parent) were strongly inter-correlated (27). Among self-reported questionnaires, only the CASP (participation) showed a moderate correlation with caregiver burden. On the other hand, burden levels were significantly associated with primary family caregiver-rated questionnaires, assessing difficulties in executive functioning, behavior, participation, quality of life, and fatigue. For an equal level of clinician-rated disability, burden was higher when the primary family caregiver reported a "negative" picture of his/her child, regardless of the specific domain concerned by this negative appreciation. This finding, together with the fact that burden was significantly associated to poorer mental health status, implies a reflection on assessing the need for psychological support to primary family caregivers over time, in order to provide adequate support, preserve mental health and reduce burden. Previous records suggest that efforts to optimize the child's environment and bolster family coping resources may enhance recovery of social problems following pediatric TBI (57).

A strong correlation was found between perceived burden and higher levels of care and needs, measured with the CANS (participants aged >15 years), but not in younger participants (<16 years, measured with the P-CANS, the pediatric adaptation of the CANS). These results are compatible with the weaker psychometric quality of the pediatric version of the CANS (compared to the adult version) (35), as well as an "age-at injury effect" on

parental burden (8,58). Age-at-injury (and therefore the degree of age-related pre-injury autonomy) could play a relevant role in determining burden. Caring for a child who has achieved regular development just up to preschool age (*e.g.* injured at 3 years) is different than looking after a child who suffered severe TBI in pre-adolescent age (*e.g.* injured at 8 years) and, as a consequence of injury, experiences a more significant involution of his/her previous abilities.

Caring for a child with chronic illness (such as cancer, chronic pain, diabetes, or TBI), may itself cause psychological vulnerability, leading to a depressive condition (59). Severe TBI is a source of considerable caregiver morbidity, even when compared with other traumatic injuries. Caregivers of children with severe TBI have been found to experience persistent stress associated with the child's injury, as well as a relative risk of clinically significant psychological symptoms nearly twice that of caregivers of children with orthopedic injuries (10).

Marsh et al. (60) assessed depression in primary caregivers following severe adult TBI. At 6-months post-injury, approximately one-third of caregivers reported clinically significant symptoms of anxiety and depression, and poor social adjustment. By 1-year post-injury, the prevalence of anxiety and depression remained the same, with one-quarter of caregivers still reporting poor social adjustment. It appears that caregivers learn some practical ways of managing the behavioral problems of individuals with TBI, and the impact of the physical impairment is relatively short-lived on the caregivers' overall quality of life. However, over time, the behavioral and cognitive problems of the individual who has undergone TBI begin to play a larger role in the level of distress experienced by the caregiver (60).

In line with the literature on families dealing with a child with intellectual disabilities (61), a significant parental concern revolves around the patient's future, related to the

perception that the patient is dependent upon the primary family caregiver. Other common caregiver reports included difficulties in meeting responsibilities for family or work, a sense that the patient requests more help than necessary, and dissatisfaction with the care job provided by the primary family caregiver. Higher perceived burden was also linked to the presence of other family members in need of caregiving and the caregiver being actively engaged in professional activities.

Adelman and colleagues (62) investigated caregiver burden related to providing support to midlife and older adults, and identified risk factors associated with higher reported caregiver burden, which included cohabitation with the patient, time spent caregiving, personal history of depression, social isolation, financial stress, and lack of choice regarding the caregiver role.

Family functioning and gender of the respondent (mother, father) were not significantly related to the perceived burden in the present study. This provides some insight into previous works that have questioned whether high levels of family burden eventually translate into global family dysfunction with increasing time since injury (63). Previous investigations (64) suggest a role for the pre-injury family environment as a predictor of family functioning after childhood TBI. Families with good communication, a positive home environment, and less pre-injury strain are more likely to have better adaptation following the injury (65). According to another report, mothers exhibit more resilience and the capability to change their habits to cope with their child's needs than fathers, who are more likely to show feelings of denial of the injury (66). In another study, parents expressed a general lack of understanding of the impact of TBI on children, particularly in case of "invisible" problems. Consequently, parents felt unsupported in coping with the child's behavioral and psychological difficulties (67). In general, parents use a variety of coping strategies, and a

higher level of denial and disengagement is associated with greater injury-related burden and distress (8).

This study has a number of limitations. The study design initially included a control group, and most self- and proxy reports of the different questionnaires were also collected in the control group (26–29,35). For the present study, the ZBI was initially proposed to the caregivers in the control group, but the items' wording did not make sense to them. Therefore, we decided to remove it from the collected data. Caregivers of control participants “only” reported on their health status (SF12), the level of care and needs of the person being cared for, and family functioning. In addition, they completed a list of questionnaires addressing their child's functioning. However, due to the significant difference in sample size between the group of participants with TBI ($n=36$) and the control group ($n=29$) with available data on the SF-12, we opted for a more conservative approach by comparing the mean scores observed in the group of participants with TBI to the mean scores expected in a standardized population. This approach was justified by preliminary analyses indicating no significant differences in the SF-12 mean scores between the control group and the standardized mean scores expected in the general population. As for family functioning and the level of care and needs, the comparisons between the group of participants with TBI and the control group have been described elsewhere (35). Although our sample is relatively large for a group of participants with severe childhood TBI compared to studies of similar nature, the sample size remains small, which might have contributed to hinder some associations.

In conclusion, 7 years after severe childhood TBI, nearly half of the primary family caregivers reported mild to moderate or moderate to severe levels of burden, and these burden levels were associated with poorer caregiver mental health status. As expected, parental burden was significantly associated with the level of overall disability, but also with worse parent-reports of patients' executive functioning, behavior, participation, quality of life and

fatigue. Furthermore, burden was significantly associated with higher levels of care and needs in older participants. The level of disability and executive functioning were the strongest independent predictors of caregiver burden. These deficits should be monitored and considered through long-term individualized follow-up, allowing for the development and implementation of timely tailored interventions focused on optimizing the child's environment and family coping resources (57). We suggest using primary family caregiver's questionnaires-reports as screening tools but basing interventions on in-depth interviews that assess the overall environmental and family situation, as well as their mood, anxiety and overall mental health. Our results emphasize the need for parents to access long-term psychological support to preserve mental health and reduce burden. Interventions targeted towards the child/adolescent's individualized goals, in a family-centered lifelong approach (capable of evolving with the contingencies and specific needs of each household), are essential to mitigate the feeling that the primary family caregiver is the only one who can provide care for the child in need. This approach is important to mitigate anxiety and worries about the child's future when the parent is no longer able to take care and, even more so, after his or her death.

Declaration of interest

The authors report there are no competing interests to declare.

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Table 1. Descriptive sociodemographic characteristics of the Primary Family Caregivers and the patients with TBI, initial injury severity, levels of care and needs, burden and 7-year post-injury concurrent outcomes

	<i>n</i>	(%)	<i>n.r.</i>	Mean	SD	Min	Max	Beyond clinical Cut-off	
	<i>n</i>	(%)						<i>n</i>	(%)
Primary Family Caregivers	36								
Age	29		7	45.45	7.87	29	64		
Gender (<i>female</i>)	25	(69)	1						
Degree of kinship	36		0						
<i>Spouse</i>	1	(3)							
<i>Mother</i>	26	(72)							
<i>Father</i>	9	(25)							
Co-residency (<i>lives in the same household</i>)	28	(78)	7						
Number of other children (<18) in the family	28		8	2.71	1.38	1	5		
Other members of the family in need of care (<i>yes</i>)	3	(8)	7						
Parental education level (<i>medium-high</i>) ^a	19	(53)	0						
Active worker (<i>yes</i>)	21	(58)	7						
Zarit Burden Interview (ZBI)	36			21.14	17.30	0	54		
Mild (0 - 20)	20	(56)							
Mild to moderate (21 - 40)	9	(25)							
Moderate to severe (41 - 60)	7	(19)							
12-item Short Form Health Survey (SF-12)									
Physical Component Score	36			46.67	10.25	25.29	62.12	5	14
Mental Component Score	36			43.56	11.20	17.15	59.38	9	25
Family Assessment Device (FAD)									
Family functioning - Total score	34			1.75	0.37	1	2.7		
Care and Needs Scale (PCANS / CANS)									
Pediatric Care and Needs Scale (<i>high level of needs</i>)	7	(35)							
Care and Needs Scale (<i>high level of needs</i>)	11	(69)							
Participants with severe TBI	36								
Age at injury (<i>years</i>)	36			7.53	4.57	0.25	14.67		
Age at follow-up (<i>years</i>)	36			15.31	4.47	7.42	22.67		
Time since injury (<i>years</i>)	36			7.78	0.85	5.92	9.33		
Gender (<i>female</i>)	14	(39)							
Preinjury education (<i>assisted and/or delayed</i>)	5	(14)							
Initial injury severity									
Lowest Glasgow Coma Scale Score (GCS)	36			5.97	1.63	3	8		
Pediatric Trauma Score (PTS)	36			3.89	2.31	-1	9		
Injury Severity Score (ISS)	36			27.58	9.79	4	50		
Length of coma (<i>days</i>)	36			6.53	4.95	1	22		

Table 1 (continued). Descriptive sociodemographic characteristics of the Primary Family Caregivers and the patients with TBI, initial injury severity, levels of care and needs, burden and 7-year post-injury concurrent outcomes

	<i>n</i>	(<i>%</i>)	<i>n.r.</i>	Mean	SD	Min	Max	Beyond clinical Cut-off	
								<i>n</i>	(<i>%</i>)
7-years post-injury concurrent outcomes									
<i>Clinician-rated outcomes</i>									
Overall disability (GOS-E/GOS-E-Peds)	36		0						
<i>Good recovery</i>	20	(56)							
<i>Moderate disability</i>	8	(22)							
<i>Severe disability</i>	8	(22)							
Motor impairments	36								
<i>Presence</i>	9	(25)							
Ongoing education	36		0						
<i>Special education</i>	12	(33)							
<i>Performance-based objective outcomes</i>									
Intellectual ability (FSIQ)	35		1	85.97	12.26	40	129	4	(11)
<i>Questionnaire-reported outcomes</i>									
Executive Functions (BRIEF)									
<i>Global Executive score (Parent-report)</i>	34		2	59.94	14.33	37	89	15	(44)
<i>Global Executive score (Self-report)</i>	11		25	61.09	11.18	46	79	3	(27)
Behavior (C/ABCL, Y/ASR)									
<i>Total behavior problems (Parent-report)</i>	33		3	59.33	10.39	39	78	14	(42)
<i>Total behavior problems (Self-report)</i>	23		13	58.09	10.50	35	75	6	(26)
Participation (CASP)									
<i>Total score (Parent-report)</i>	36		0	88.03	11.65	57.5	100	18	(50)
<i>Total score (Self-report)</i>	26		10	89.47	10.61	62.5	100	5	(19)
Quality of Life (PedsQL)									
<i>Total score (Parent-report)</i>	25		11	71.68	18.89	34.8	96	9	(36)
<i>Total score (Self-report)</i>	32		4	70.67	18.49	34.8	98.9	10	(31)
Fatigue (MFS PedsQL)									
<i>Total score (Parent-report)</i>	25		11	67.78	20.34	31.9	100	16	(64)
<i>Total score (Self-report)</i>	32		4	60.63	18.11	27.8	90.3	17	(53)

TBI: Traumatic Brain Injury; ^a At least one of the parents had graduated from high school; *n.r.*: number of non-respondents; SF-12: 12-item Short Form Health Survey [M (SD)=50 (10), cut-off (< 1.5 SD) < 70](33); GOS-E / GOS-E-Peds: Glasgow Outcome Scale-Extended / Glasgow Outcome Scale-Extended, Pediatric version; FSIQ: Full Scale Intellectual Quotient [M(SD)=100 (15), cut-off (< 2 SD) < 70](40); BRIEF: Behavior Rating Inventory of Executive Function [M(SD)=50 (10), clinical cut-off (≥ 1.5 SD): ≥ 65](41,42); C/ABCL: Child/Adult Behavior; Y/ASR: Youth/Adult Self Report Checklist [M(SD)=50(10), cut-off (≥ 1.5 SD): ≥ 64](43,44); CASP: Child and Adolescent Scale of Participation inventory [range: 0 to 100; cut-off (10th percentile): parent-report ≤ 88.9 , self-report ≤ 78.8](68); PedsQL: Pediatric Quality of Life inventory [M(SD) parent-report=81.3 (15.9), self-report=82.9 (13.2); cut-off (<1SD): parent reports ≤ 65.4 ; self-reports ≤ 69.7](45,46,69,70); MFS: Multidimensional Fatigue Scale [M(SD) parent-report=87.6 (11.4), self-report=77.4 (15.3); cut-off (<1SD: parent report ≤ 76.2 ; self-report ≤ 62.1)](27).

Table 2. Associations of the Zarit Burden Interview. Univariate analysis: Student’s t-test or Pearson

	7-years post-severe TBI Zarit Burden Interview			
	<i>n</i>	<i>t</i>	<i>r</i>	<i>p</i>
Primary Family Caregiver				
Others members of the family in need of care	29			0.017
Short Form 12 Questionnaire (SF-12)				
Physical Component Score (PCS 12)	36		-0.12	0.48
Mental Component Score (MCS12)	36		-0.58	0.0002
Family Assessment Device (FAD)				
Family functioning - Total score	34		0.20	0.259
Initial injury severity				
Lowest Glasgow Coma Scale Score	36		-0.14	0.39
Pediatric Trauma Score	36		0.16	0.35
Injury Severity Score	36		0.37	0.03
Length of coma (<i>days</i>)	36		0.33	0.05
7-year post-injury outcomes				
Clinician-rated outcomes				
Overall disability (GOS-E / GOS-E-Peds)	36		0.56	0.0003
Motor deficits	36	-0.50		0.62
Care and Needs Scale				
Pediatric Care and Needs Scale (PCANS)	20		0.27	0.245
Care and Needs Scale (CANS)	16		0.82	0.0001
Ongoing education	36	-1.33		0.19
Performance-based outcomes				
Intellectual ability (FSIQ)	35		-0.17	0.32
Questionnaire-reported outcomes				
Executive Functions (BRIEF)				
Global Executive score (Parent-report)	34		0.70	<0.0001
Global Executive score (Self-report)	11		0.14	0.68
Behavior (C/ABCL, Y/ASR)				
Total behavior problems (Parent-report)	33		0.64	<0.0001
Total behavior problems (Self-report)	23		0.39	0.06
Participation (CASP)				
Total score (Parent-report)	36		-0.60	0.0001
Total score (Self-report)	26		-0.54	0.005
Quality of Life (PedsQL)				
Total score (Parent-report)	25		-0.61	0.001
Total score (Self-report)	32		-0.26	0.156
Fatigue (MFS PedsQL)				
Total score (Parent-report)	25		-0.72	<0.0001
Total score (Self-report)	32		-0.23	0.204

t: Student’s *t*-test; *r*: Pearson correlation coefficient; *r*[†]: Partial correlation coefficients adjusted for overall disability assessed with the Glasgow Outcome Scale-Extended (GOS-E) or Glasgow Outcome Scale-Extended, Pediatric version (GOS-E-Peds).

Multivariate analyses: ISS and length of coma no more significant when overall disability (GOS) is considered. Stepwise backwards selection: GOS and BRIEF-parent report remained significant.

Table 3. Correlation and partial correlation matrix (GOS-E-adjusted) of the Zarit Burden Interview, the GOS-E, the SF12-MCS, and the parent-reported assessments 7-years post-injury

	GOS-E			SF12			BRIEF			C/ABCL			CASP			PedsQL			MFS		
	GOS-E-Peds			MCS												PedsQL					
	<i>n</i>	<i>r</i>	<i>r</i> [†]	<i>n</i>	<i>r</i>	<i>r</i> [†]	<i>n</i>	<i>r</i>	<i>r</i> [†]	<i>n</i>	<i>r</i>	<i>r</i> [†]	<i>n</i>	<i>r</i>	<i>r</i> [†]	<i>n</i>	<i>r</i>	<i>r</i> [†]	<i>n</i>	<i>r</i>	<i>r</i> [†]
ZBI	36	0.56***	-	36	-0.58***	-0.49**	34	0.70***	0.67***	33	0.64***	0.55**	36	-0.60***	-0.49**	25	-0.61***	-0.53**	25	-0.72***	-0.59**
GOS-E/GOS-E-Peds				36	-0.34*	-	34	0.33	-	33	0.39*	-	36	-0.41*	-	25	-0.38	-	25	-0.54**	-
SF12-MCS				-	-	-	34	-0.48**	-0.40*	33	-0.25	-0.10	36	0.34*	0.23	25	0.28	0.20	25	0.44*	0.36
BRIEF							-	-	-	33	0.85***	0.83***	34	-0.69***	-0.64***	23	-0.53**	-0.43*	23	-0.60**	-0.50**
C/ABCL										-	-	-	33	-0.74***	-0.69***	22	-0.67***	-0.58**	22	-0.64**	-0.52*
CASP													-	-	-	25	0.52**	0.43*	25	0.61**	0.49*
PedsQL																-	-	-	25	0.80***	0.77***

r: Pearson correlation coefficient; *r*[†]: Partial correlation coefficients adjusted for overall disability (GOS-E / GOS-E-Peds); * *p*<0.05, ** *p*<0.01, *** *p*<0.001; ZBI: Zarit Burden Interview; GOS-E/GOS-E Peds: Overall disability assessed with the Glasgow Outcome Scale-Extended (GOS-E) or Glasgow Outcome Scale-Extended, Pediatric version (GOS-E-Peds). SF12-MCS: Mental component Score assessed with the Short Form 12 Questionnaire (SF-12). BRIEF: Executive Functions assessed with the Behavior Rating Inventory of Executive Function (BRIEF). C/ABCL: Behavior assessed with the Child/Adult Behavior Checklist (C/ABCL). CASP: Participation assessed with the Child and Adolescent Scale of Participation (CASP). PedsQL: Quality of Life assessed with the Pediatric Quality of Life inventory (PedsQL). MFS PedsQL: Fatigue assessed with the Multidimensional Fatigue Scale (PedsQL MFS).

Table 4. Regression analysis of the Zarit Burden Interview

	Zarit Burden Interview			
	R^2	β	$S(\beta)$	p
Step 1	0.69			
GOS-E / GOS-E-Peds		3.94	2.16	0.09
SF12-MCS		-0.47	0.34	0.19
BRIEF		0.28	0.42	0.52
C/ABCL		0.07	0.62	0.91
CASP		0.009	0.31	0.97
PedsQL		-0.12	0.25	0.65
MFS		-0.04	0.26	0.87
Final step	0.62			
GOS-E / GOS-E-Peds		5.14	1.74	0.008
BRIEF		0.55	0.18	0.006

GOS-E/GOS-E-Peds: Glasgow Outcome Scale-Extended or Glasgow Outcome Scale-Extended, Pediatric version.

SF12-MCS: Short Form 12 Questionnaire Mental Component Score. BRIEF: Behavior Rating Inventory of Executive

Function. C/ABCL: Child/Adult Behavior Checklist. CASP: Child and Adolescent Scale of Participation. PedsQL:

Pediatric Quality of Life inventory. MFS: Multidimensional Fatigue Scale.

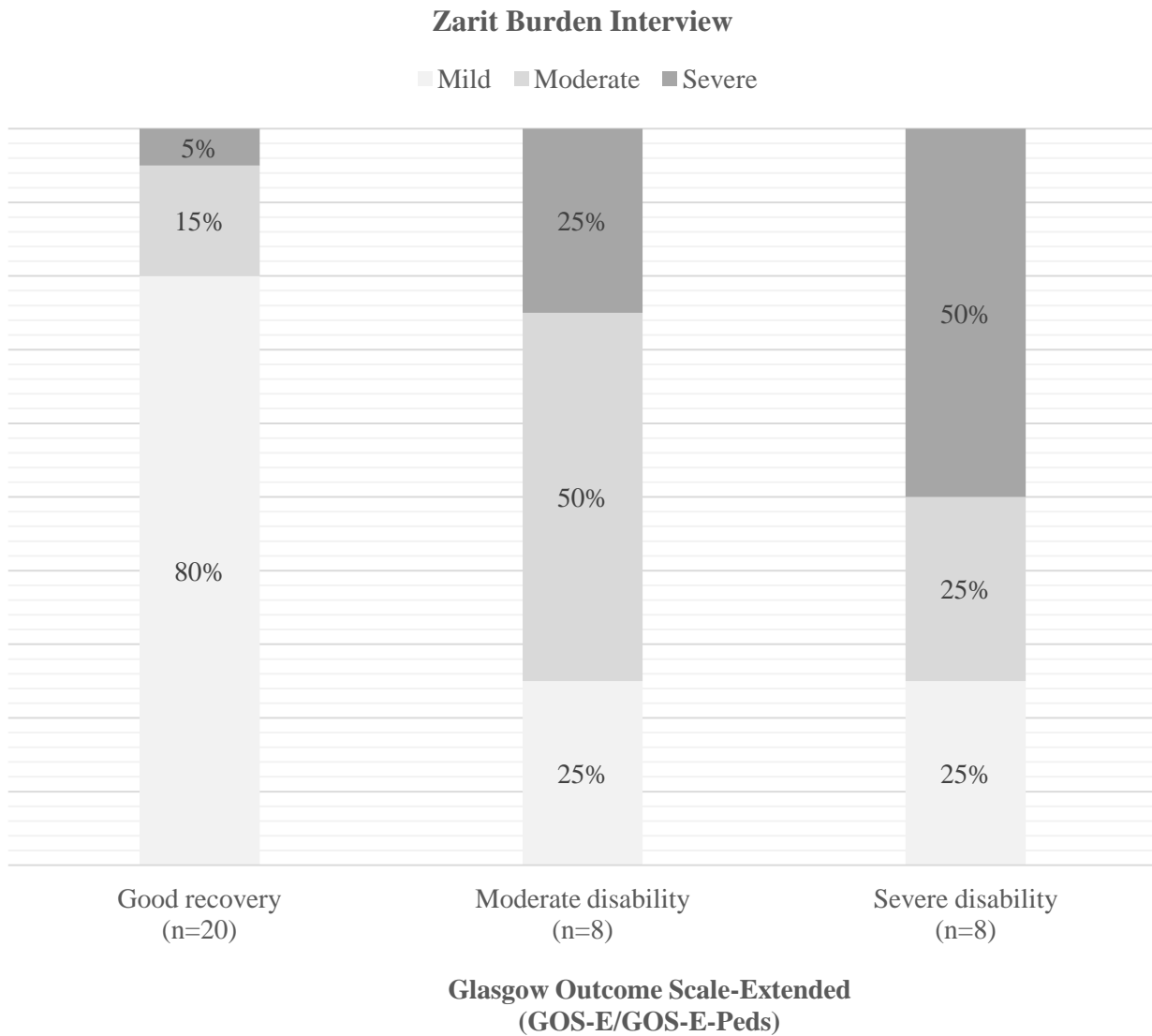


Figure 1. Percentages of mild, mild to moderate and moderate to severe burden assessed with the Zarit Burden Interview ($n=36$) according to Glasgow Outcome Scale-Extended outcomes.

(Chi-square $(4, 32) = 13.75, p = 0.008$).

SUPPLEMENTARY MATERIAL

Table s1. Rank order of the 22-items' mean values from the Zarit Burden Interview observed in the group of participants with severe Traumatic Brain Injury

Item	Zarit Burden Interview (<i>n</i> =36)	Mean	SD	Min	Max
7.	I am afraid of what the future holds for the person I care for.	2.44	1.56	0	4
8.	I feel he/she is dependent upon me.	1.69	1.77	0	4
3.	I feel stressed between caring for him/her and trying to meet other responsibilities for my family or work.	1.47	1.58	0	4
1.	I feel that the person I care for asks for more help than he/she needs.	1.42	1.40	0	4
21.	I feel I could do a better job in caring for him/her.	1.39	1.34	0	4
20.	I feel I should be doing more for him/her.	1.17	1.38	0	4
2.	Because of the time I spend with him/her, I do not have enough time for myself.	1.14	1.55	0	4
12.	I feel that my social life has suffered because I am caring for this person.	1.11	1.43	0	4
17.	I feel that I have lost control of my own life since this person's illness.	1.06	1.41	0	4
22.	Overall, how burdened do you feel in caring for this person?	1.03	1.44	0	4
14.	I feel that this person expects me to take care of him/her as if I was the only one he/she could depend on.	1.00	1.31	0	4
4.	I feel embarrassed over his/her behavior.	0.94	1.26	0	4
10.	I feel that my health has suffered because of my involvement with the person I care for.	0.94	1.41	0	4
15.	I feel that I do not have enough money to support this person in addition to the rest of our expenses.	0.86	1.17	0	4
11.	I feel that I do not have as much privacy as I would like because of the person I care for.	0.61	1.25	0	4
16.	I feel that I will be unable to take care of him/her much longer.	0.56	1.03	0	3
9.	I feel strained when I am around the person I care for.	0.53	0.81	0	3
6.	I feel that he/she currently affects my relationship with other family members or friends in a negative way.	0.50	0.74	0	2
5.	I feel angry when I am around the person I care for.	0.43	1.04	0	4
19.	I feel uncertain about what to do about the person I care for.	0.42	1.02	0	4
13.	I feel uncomfortable about having friends over because of him/her.	0.25	0.60	0	2
18.	I wish I could leave the care of this person to someone else.	0.22	0.59	0	2
Averaged total score		0.96	0.79	0	2.45

SUPPLEMENTARY MATERIAL

Total score	21.14	17.30	0	54
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Each item from the Zarit Burden Interview is scored in a 5-point scale (0=Never; 1=Rarely; 2=Sometimes; 3=Quite frequently; 4=Nearly always).

SUPPLEMENTARY MATERIAL

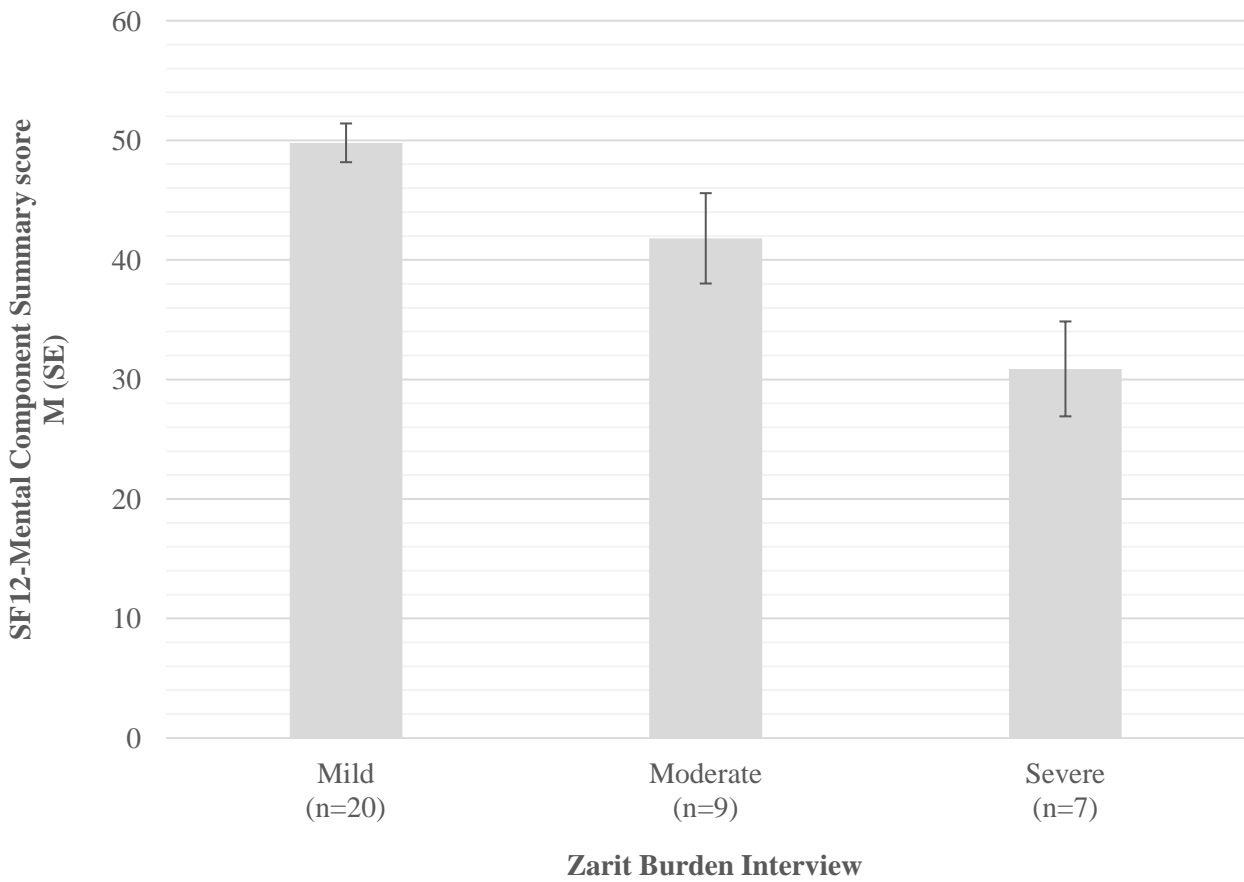


Figure s1. Distribution of the mean scores and standard error means of the mental health status assessed with the SF12-Mental Component Summary score ($n=36$) according to the Primary Family Caregiver's reported burden evaluated with the Zarit Burden Interview.

SUPPLEMENTARY MATERIAL

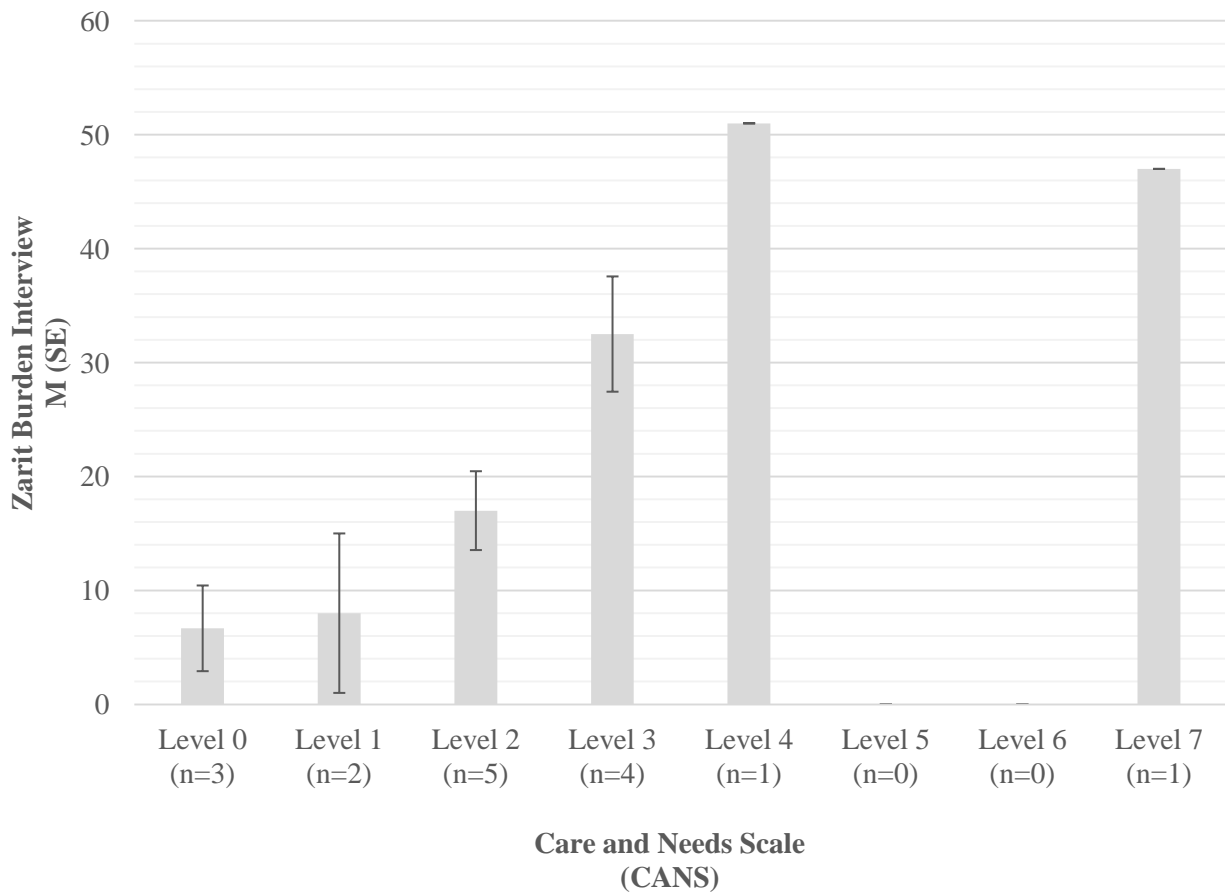


Figure s2. Distribution of the mean scores and standard error means of the Zarit Burden Interview ($n=16$) according to the level of needs assessed by the Care and Needs Scale (age ≥ 16 years) in participants with severe Traumatic Brain Injury 7-years post-injury.

Lower scores indicate increased level of care and needs.

Level 0 = Can live in the community, totally independently

Level 1 = Can live alone, but needs intermittent (i.e., less than weekly) support

Level 2 = Can be left alone for almost all week

Level 3 = Can be left alone for a few days a week

Level 4 = Can be left alone for part of the day and overnight

Level 5 = Can be left alone for part of the day, but not overnight

Level 6 = Can be left alone for a few hours

Level 7 = Cannot be left alone